

Please cite the Published Version

Harding, Sam (2019) Psychological understanding of the impact of health interventions in relation to chronic conditions. Doctoral thesis (PhD), Manchester Metropolitan University.

Downloaded from: <https://e-space.mmu.ac.uk/626262/>

Usage rights:  [Creative Commons: Attribution-Noncommercial-No Derivative Works 4.0](#)

Additional Information: A thesis submitted in fulfilment of the requirements of Manchester Metropolitan University (MMU), for the degree of Doctor of Philosophy by Published Work (Route 2).

Enquiries:

If you have questions about this document, contact openresearch@mmu.ac.uk. Please include the URL of the record in e-space. If you believe that your, or a third party's rights have been compromised through this document please see our Take Down policy (available from <https://www.mmu.ac.uk/library/using-the-library/policies-and-guidelines>)

Psychological understanding of the impact of
health interventions in relation to chronic
conditions.

S A Harding
PhD 2019

Psychological understanding of the impact of health interventions in relation to chronic conditions.

Sam A Harding

A thesis submitted in fulfilment of the requirements of Manchester Metropolitan University (MMU), for the degree of Doctor of Philosophy by

Published Work (Route 2)

Faculty of Health, Psychology and Social Care, Manchester Metropolitan University (MMU), Manchester.

2019

Contents

Contents	2
Table of tables	6
Table of figures	7
List of abbreviations	8
Abstract	9
Dedication	11
Acknowledgements	11
Chapter 1 Introduction	12
1.1 Aims of the thesis	12
1.2 The structure of the thesis	13
1.3 The papers presented in this thesis	16
Chapter 2 Philosophy	18
2.1 Philosophical basis for the research	18
2.1.1 Ontology	18
2.1.2 Epistemology	19
2.1.3 Axiology	19
2.1.4 Rhetoric	20
2.1.5 Methodology	20
2.1.6 Philosophical conclusion	20
2.2 Autoethnography	21
2.2.1 Researchers auto-ethnographic stance	21
2.3 Summary	22
Chapter 3 Single Time Point Studies	23
3.1 Paper 1 - Clinical experience leading to the research work and production of the paper	23
3.1.1 Research Questions	24
3.1.2 Study Design	24
3.1.3 The prevalence of PTSD and changes in PTSD symptoms following pulmonary rehabilitation.	25
3.1.4 Study limitations	34
3.1.5 Contribution to knowledge and autoethnographic issues	35
3.1.7 Future research questions	37
3.2 Paper 2 - Clinical experience leading to the research work and production of the paper	38
3.2.1 Research Question	39
3.2.2 Study Design	39
3.2.3 The impact of treatment for head and neck cancer on positive psychological change within a year of completing treatment.	42
3.2.4 Study limitations	49
3.2.5 Contribution to knowledge and autoethnographic issues	52
3.2.6. Presenting findings to an HNC population	52
3.2.7 Future research questions	54
3.3 Summary	54

3.3.1 Summary of contribution to knowledge	54
3.3.2 Summary of autoethnographic issues	54
Chapter 4 Pre & Post Studies	55
4.1 Paper 3 - Clinical experience leading to the research work and production of the paper	55
4.1.1 Research Questions.....	56
4.1.2 Study Design.....	56
4.1.3 Educational impact of pulmonary rehabilitation: Lung Information Needs Questionnaire	57
4.1.4 Study Limitations.....	65
4.1.5 Contribution to knowledge and autoethnographic issues	65
4.1.7 Future research questions	67
4.2 Paper 4- Clinical experience leading to the research work and production of the paper	68
4.2.1 Research Questions.....	68
4.2.2 Study Design.....	68
4.2.3 Impact of perioperative hyperbaric oxygen therapy on the quality of life of maxillofacial patients who undergo surgery in irradiated fields	69
4.2.4 Study Limitations.....	78
4.2.5 Contribution to knowledge and autoethnographic issues	79
4.2.6 Future research questions	81
4.3 Paper 5 - Clinical experience leading to the research work and production of the paper	82
4.3.1 Research Questions.....	83
4.3.2 Study Design.....	83
4.3.3 Effects of Hyperbaric Oxygen Therapy on Quality of Life in Maxillofacial Patients with Type III Osteoradionecrosis	83
4.3.4 Study Limitations.....	91
4.3.5 Contribution to knowledge and autoethnographic issues	91
4.3.6 Informing the research journey	92
4.4 Summary	93
4.4.1 Summary of contribution to knowledge	93
4.4.2 Summary of the autoethnographic issues	94
Chapter 5 Longitudinal Studies	95
5.1 Paper 6 - Clinical experience leading to the research work and production of the paper	95
5.1.1 Research Questions.....	95
5.1.2 Study Design.....	96
5.1.3 The trajectory of positive psychological change in a head and neck cancer population	97
5.1.4 Study Strengths and Limitations	105
5.1.5 Contribution to knowledge and autoethnographic issues	108

5.1.6 Future research questions	110
5.2 Summary	111
5.2.1 Summary of contribution to knowledge	111
5.2.2 Summary of autoethnographic issues	111
Chapter 6 Systematic Reviews	113
6.1 Paper 7 - Clinical experience leading to the research work and production of the paper	113
6.1.1 Research Questions.....	114
6.1.2 Study Design.....	114
6.1.3 Existence of benefit finding and posttraumatic growth in people treated for head and neck cancer: a systematic review.....	115
6.1.4 Study Limitations.....	139
6.1.5 Contribution to knowledge and autoethnographic issues	141
6.1.6 Future research questions	142
6.2 Paper 8 - Clinical experience leading to the research work and production of the paper	143
6.2.1 Research Questions.....	143
6.2.2 Study Design.....	143
6.2.3 Characteristics of Parent-Child Interactions: A systematic review of studies comparing children with primary language impairment and their typically developing peers	143
6.2.4 Study Limitations.....	156
6.2.5 Contribution to knowledge and autoethnographic issues	157
6.2.6 Future research questions	158
6.3 Paper 9 - Clinical experience leading to the research work and production of the paper	160
6.3.1 Research Questions.....	160
6.3.2 Study Design.....	161
6.3.3 A systematic review and classification of interventions for speech-sound disorder in preschool children	162
6.3.4 Study Limitations.....	185
6.3.5 Contribution to knowledge and autoethnographic issues	185
6.3.6 Future research questions	187
6.4 Summary	188
6.4.1 Summary of contribution to knowledge	188
6.4.2 Summary of autoethnographic issues	189
Chapter 7 Discussion of Research Journey	190
7.1 Research apprenticeship	190
7.1.1 Research methods skills	191
7.1.2 Discipline - specific skills	191
7.1.3 Personal skills	191
7.1.4 Range of setting and cross-professional working	192
7.2 Contribution to new knowledge	192

7.2.1 Contribution to knowledge of included single time point studies (Chapter 3)	192
7.2.2 Contribution to knowledge of pre, post intervention studies (Chapter 4)	192
7.2.3 Contribution to knowledge of longitudinal studies (Chapter 5)	193
7.2.4 Contribution to knowledge of systematic reviews (Chapter 6)	193
7.2.5 Summary of contribution to our psychological understanding of the impact of health interventions in relation to chronic conditions	194
7.3 Future direction of my research	195
7.3.1 Changing face of research - Greater importance of PPI and making research directly relevant	195
7.4 Now bidding as a Chief Investigator	196
7.4.1 Novel health condition - Male Breast Cancer	196
7.4.2 Novel methodologies – Drawing as a novel methodology for use with people experiencing HNC.....	196
7.4.3 Parent Child Interaction	197
7.5 Conclusion	197
References.....	199
Appendix 1: Contribution to publication forms	210
Appendix 2: Impact of Research – Paper 1	219
Appendix 3: Paper 1 poster presentation	222
Appendix 4: Description of questionnaires used in paper 2	223
Appendix 5: Impact of research of paper 2	226
Appendix 6: Paper 2 Poster presentation	229
Appendix 7: Impact of research of paper 3	230
Appendix 8: LINQ Poster	235
Appendix 9: Impact of research of paper 4	236
Appendix 10: Hyperbaric Oxygen Therapy and Quality of Life Poster	241
Appendix 11: Hyperbaric Oxygen Therapy and Quality of Life Presentation	242
Appendix 12: Impact of research of paper 5	247
Appendix 13: Impact of research of paper 6	250
Appendix 14: Paper 6 Oral presentation	252
Appendix 15: Mediators and Moderators of PPC	262
Appendix 16 Measure of Positive Psychological Change	265
.....	265
Appendix 17: Impact of research of paper 7	274
Appendix 18: Impact of research of paper 8	278
Appendix 19: Impact of research reported of paper 9	281
Appendix 20: Certificate of recognition	284
Appendix 21: Southmead Hospital Charity, Expression of Interest – Male Breast Cancer	285
Appendix 22: Drawing as a novel methodology – Harding and Bradford (2019)	292
Appendix 23: Heather van der Lely Foundation Trust Project Summary	302

Table of tables

Table 1: interventions against the patient cohorts represented in the selected papers17

Table of figures

Figure 1: Data collection points	78
Figure 2: Severe deformity	83
Figure 3: There is an I in team	111
Figure 4: Numbers of studies from the systematic review that were allocated to each theme.....	162
Figure 5: Defining the Role of Authors	188

List of abbreviations

Abbreviation	Meaning
COPD	Chronic Obstructive Pulmonary Disease
DDRC	Diving Diseases Research Centre
HBOT	Hyperbaric oxygen therapy
HNC	Head and Neck Cancer
HRQoL	Health Related Quality of Life
LINQ	Lung Information Needs Questionnaire
MCID	Minimal Clinically Important Difference
MCS	Mental Component Summary
ORN	Osteoradionecrosis
PCIT	Parent Child Interaction Therapy
PCS	Physical Component Summary
Peri	During
PICO	Population, Intervention, Comparison, Outcome
Post	After
PPC	Positive Psychological Change
PPI	Public Patient Involvement
PR	Pulmonary Rehabilitation
Pre	Before
PROMs	Patient Reported Outcome Measures
PTGI	Post-traumatic growth inventory
PTSD	Post Traumatic Stress Disorder
QoL	Quality of Life
SF-12	Medical Outcomes Short Form - 12
SLQ	Silver Lining Questionnaire
SSD	Speech Sound Disorders
UoW	University of Washington

Abstract

This thesis describes original research by the author into the quality of life impact of significant health interventions on patients' psychology. Nine publications spanning 10 years of research, multiple medical specialities, and study methodologies, are presented. A unifying factor across this research is that all participants have been in receipt of an evidence-based intervention to address their specific challenges.

Six papers describe primary data collection and analysis to gain an understanding of prevalence or change over time of a health psychology phenomenon. Following on from these papers, three systematic reviews are included. These present the evidence base for psychological phenomena in a population, or the effectiveness of an intervention.

The thesis is structured around the design of the studies included. Three distinct study designs are employed; 2 single time point or 'snapshot' paper, 3 two-time point or 'Pre- & Post' studies, and a longitudinal study. All adopted a cohort approach for both methodological and practical reasons. Each produced findings that contribute to our knowledge of the phenomenon under investigation, as outlined in the following chapters.

The longitudinal cohort study, additionally contributed methodologically to the field of health psychology research. By using a novel adaptation of cross-sequential design, and Linear Mixed-effect Modelling for analysis, this research demonstrated how long term conditions in relatively small populations can be rigorously investigated.

All six primary research papers recognise the importance of Public Patient Involvement. I have always taken a strong moral stance on the inclusion of the patient perspective in study design and data interpretation, and it is now being formally included as a core part of health psychology research design and funding. As a Health Psychologist and methodologist, this thesis reflects on how working with research participants across projects has influenced my clinical and research practice, and how I have tried to explore the impact of their treatments on psychological well-being.

I present how each paper has contributed to new knowledge and how it has informed my development as an independent researcher. Lastly, I propose a

research agenda, informed by the research in this thesis, with suggestions of how this may fit with my research interests, the changing face of research and service provision within the NHS and social care.

Dedication

Dedicated to Dr John Bradford, who has put up with my chasing knowledge and experience for more years than I am sure he wishes to recall.

Acknowledgements

Many thanks to my advisor Prof Juliet Goldbart for suggesting that this programme of work was possible, and subsequently her tireless advice and support throughout the process. I am also indebted to my colleagues at the Bristol Speech and Language Therapy Research Unit, and Research and Innovation department at North Bristol NHS Trust, for their support during the completion of this thesis.

I would also like to thank all my co-authors and researchers for allowing me to work with them and to use our joint publications in this thesis.

Chapter 1 Introduction

The purpose of this thesis is to present nine publications for which I am the sole or joint author, and to demonstrate how these publications contribute to understanding the psychological impact of the disease and/or the treatment being experienced. In particular, the focus of a number of the papers is quality of life (QoL). All papers relate to health interventions addressing a range of chronic conditions. The papers are grouped according to their study design.

1.1 Aims of the thesis

The aim of the thesis is to describe my research into the impact of health interventions on patients' psychology with emphasis on QoL, across positive and negative psychological change. What unites this research is that the participants have all experienced a significant chronic health-related challenge, and all have been in receipt of an evidence-based intervention to address this challenge. The interventions included are:

- Hyperbaric oxygen therapy (HBOT)
- Treatments for cancer including surgery, radiotherapy, chemotherapy and combined therapies
- Pulmonary rehabilitation (PR)
- Parent Child Interaction Therapy (PCIT)

The patient cohorts that will be covered by these treatments are:

- People who had undergone treatment for Head and Neck Cancer (HNC)
- People who had been diagnosed with Chronic Obstructive Pulmonary Disease (COPD)
- Children with Developmental Speech or Language difficulties, specifically those with a speech sound disorder

A chronic condition is defined by Holman and Lorig (2000) as; usually having a gradual onset, being of indefinite duration, usually with multiple changing causes over time, having an uncertain diagnosis or prognosis, having an uncertain trajectory, and, where health care professionals and patients have complementary knowledge.

Head and neck cancer and COPD easily fit into this definition, but speech sound disorder need greater consideration.

Speech sound disorder onset is gradual in children in as far as they are not following a normal development profile. Shriberg (2010) estimates that as many as 75% of children who present with SSD at age 3 will have normalised speech by age 6.

However, a substantial minority of children have persistent speech disorder which continues into older childhood and sometimes adulthood, thus it can be of indefinite duration. Persistent speech disorder can be observed in children who have no identifiable cause for their difficulties. Wren et al (2016) found that 3.6% of children in a large scale community population study had persistent speech disorder at age 8. Thus for this group of children speech sound disorder can be seen as a chronic condition.

1.2 The structure of the thesis

Within the domain of health psychology, my particular contribution has been methodological. I am a methodologist.

Each paper makes individual contributions to our understanding of how the psychology of a person can be impacted by their treatment for a health condition. These impacts may manifest as a positive or negative reframing of the participant's world view, or with specific changes in elements of QoL. The sophistication of our understanding is dependent on our methodology, and the papers are grouped by research design to demonstrate this.

I also show that how a phenomenon is investigated (the methodology) can affect the clinical team's understanding of the patient impact. Drawing this research together allows the reader to view my wider contribution to our understanding of psychological change. This thesis also allows me (the researcher) to reflect and more fully ground myself within my personal ethnography and discuss how this lens affects my past choices, and future plans.

The overall structure of the thesis is as follows:

Chapter 2 describes my philosophical position and presents my auto-ethnographical stance, through my reflection of working personally with research participants, co-researchers, and clinicians.

Chapters 3 to 6 are structured around the featured papers, with each chapter presenting a distinct methodology. They will demonstrate how I have been influenced by patients' experiences and subsequently tried to explore the impact of their treatments on psychological well-being. I will discuss the limitations of the methods used and how these affect the interpretation of the data within the publication and its usefulness to the practice of clinicians and allied health professionals.

Chapter 3 presents two papers. The first paper investigates the prevalence of psychological problems in a population with COPD. The second paper looks at the impact of treatment for HNC in the short term. Both papers are single time point studies.

1. Jones RCM, **Harding SA**, Chung M, Campbell J (2009) The prevalence of PTSD and changes in PTSD symptoms following pulmonary rehabilitation. *Journal of Cardiopulmonary Rehabilitation and Prevention. Journal of Cardiopulmonary Rehabilitation and Prevention*. 29(1): 49-56. DOI: 10.1097/HCR.0b013e318192787e.
2. **Harding S** & Moss, T (2018) The impact of treatment for head and neck cancer on positive psychological change within a year of completing treatment. *International journal of oral and maxillofacial surgery*. DOI: 10.1016/j.ijom.2017.07.023

Chapter 4 presents three papers containing data from patients before (pre) and after (post) intervention or therapy. The first paper in this chapter presents findings on the impact of pulmonary rehabilitation on people with COPD. The other two papers look at the impact of treatment for people that have had HNC.

3. Jones RMC, Wang X, **Harding SA**, Bott J & Hyland M (2008) Educational impact of pulmonary rehabilitation: Lung Information Needs Questionnaire. *Respiratory Medicine*. 102(10): 1439-45.
4. **Harding SA**, Courtney DJ, Hodder SC, Bryson PJ (2008) Impact of perioperative hyperbaric oxygen therapy on the quality of life of maxillofacial patients who undergo surgery on irradiated fields. *International Journal of Oral & Maxillofacial Surgery*. 37(7): 617-624. DOI: 10.1016/j.ijom.2008.04.004.

5. **Harding SA**, Hodder SC, Courtney DJ, Bryson PJ (2012) The Effects of Hyperbaric Oxygen Therapy on Quality Of Life in Maxillofacial Patients with Type III Osteoradionecrosis. *Journal of Oral & Maxillofacial Surgery*. **70**(12):2786-92. DOI: 10.1016/j.joms.2012.04.011.

Chapter 5 presents one paper and focuses on the longitudinal impact of chronic diseases and predictors of psychological changes.

6. **Harding S** (2018) The trajectory of positive psychological change in a head and neck cancer population. *International journal of oral and maxillofacial surgery*. DOI: 10.1016/j.ijom.2017.09.010

Chapter 6 presents three systematic reviews, where evaluation of published evidence allows a clear understanding of the information available to clinicians. I will discuss how undertaking systematic reviews has informed my practice. I will reflect on some of the challenges associated with this type of review such as only using published or readily available studies, and how this might affect clinical decision-making.

7. **Harding S**, Sanipour F & Moss T (2014) Existence of benefit finding and posttraumatic growth in people treated for head and neck cancer: a systematic review. *PeerJ* <https://peerj.com/articles/256.pdf>
8. Blackwell AKM, **Harding SA**, Babayigit S & Roulstone S (2014) Characteristics of Parent-Child Interactions: A Systematic Review of studies comparing children with primary language impairment and their typically developing peers. *Communication Disorder Quarterly*, DOI: 10.1177/1525740114540202
9. Wren Y, **Harding S**, Goldbart J & Roulstone S (2018) A systematic review and classification of interventions for speech-sound disorder in preschool children. *International Journal of Language & Communication Disorders*. DOI: 10.1111/1460-6984.12371

Chapter 7 draws the implications of the preceding chapters together. It goes on to propose a research agenda that follows on from my published works and how this may fit the changing face of research and service provision within the NHS and social care.

The percentage contributions made by each of the researchers on their respective papers are given on the 'Contribution to publications' forms in Appendix 1.

1.3 The papers presented in this thesis

The papers included in this thesis relate to four main interventions and three patient cohorts. The papers will be discussed in relation to the methodology used and how this affects the interpretation, and generalisability of the findings.

The publications included use cohorts, to gain an understanding of prevalence, or change over time. Several systematic reviews are included and these show the evidence base for psychological phenomena in a population. Systematic reviews can also suggest the effectiveness of an intervention. Table 1 shows the interventions against the patient cohorts represented in the selected papers. The nine papers form the core of this thesis and these and will be referred to throughout. My percentage contribution for each of the papers is presented in Table 1 and confirmed in the documentation included in Appendix 1.

Table 1: interventions against the patient cohorts represented in the selected papers

Paper Number	Paper Title	Chapter Number	Year of Publication	Patient Conditions	Intervention	Percentage Contribution
1	The prevalence of posttraumatic stress disorder in patients undergoing pulmonary rehabilitation and changes in PTSD symptoms following rehabilitation.	3	2009	Chronic Obstructive Pulmonary Disease	Pulmonary Rehabilitation	30%
2	The impact of treatment for head and neck cancer on positive psychological change within a year of completing treatment	3	2018	Head and Neck Cancer	Treatment for Head and Neck Cancer	95%
3	Educational impact of pulmonary rehabilitation: Lung Information Needs Questionnaire	4	2008	Chronic Obstructive Pulmonary Disease	Pulmonary Rehabilitation	25%
4	Impact of perioperative hyperbaric oxygen therapy on the quality of life of maxillofacial patients who undergo surgery in irradiated fields	4	2008	Head and Neck Cancer	Hyperbaric Oxygen Therapy	85%
5	Effects of hyperbaric oxygen therapy on quality of life in maxillofacial patients with type III Osteoradionecrosis	4	2012	Head and Neck Cancer	Hyperbaric Oxygen Therapy	85%
6	The trajectory of positive psychological change in a head and neck cancer population	5	2018	Head and Neck Cancer	Treatment for Head and Neck Cancer	100%
7	Existence of benefit finding and posttraumatic growth in people treated for head and neck cancer: a systematic review	6	2014	Head and Neck Cancer	-	85%
8	Characteristics of parent child interactions: A systematic review of studies comparing children with primary language impairment and their typically developing peers.	6	2014	Language Impairment	-	10%
9	A systematic review and classification of interventions for speech-sound disorder in preschool children	6	2018	Speech Sound Disorder	-	60%

Chapter 2 Philosophy

This chapter describes my (the researcher) philosophical position and presents my auto-ethnographical stance, through my reflection on working with research participants, co-researchers and clinicians.

2.1 Philosophical basis for the research

There is a need to discuss philosophy since it has a fundamental impact on the research conducted, the results derived and the solution developed. Creswell (1994) identifies five levels of assumptions regarding research in general. These assumptions relate to the ontological, epistemological, axiological, rhetorical and methodological positions that researchers adopt when considering their research and the questions that they are seeking to answer. The most fundamental of these assumptions is the ontological one since this deals with seeking to define what is meant by 'reality' and the position of the researcher within that reality. For this reason, it will be considered in some depth; the other assumptions follow on from this initial position and the purpose of their inclusion here is to demonstrate an understanding of the issues raised and to ensure that a consistent philosophical thread runs through the research.

2.1.1 Ontology

Ontology relates to the branch of metaphysics concerned with the nature of being, that is the degree to which there is an absolute reality that is distinguishable from the observer's perception (Creswell, 1994). At one end of the ontological spectrum, there is the existential opinion that there is no absolute reality, that what we know as reality is merely a construct formed by our brains to interpret the signals received from the senses. There is no method for independently verifying those signals and so there is no method for independently verifying reality. In a similar vein, the causal relationships observed are generated by the brain to interpret better the signals received and may not reflect any absolute laws. At the extreme, there can be no independent verification for the existence of others, leading to the solipsist stance that everything, including the existence of others, is a construct of the brain. This has profound implications for research since any knowledge acquired will be rooted in the constructs of the researcher. There is thus no way of transferring those

constructs to another person and no way of generalising the knowledge gained (Creswell, 1994).

The axiomatic realist approach (Meredith *et al.*, 1989) at the other end of the continuum suggests that there is a rational, independent reality and that we all experience the same reality (Sears *et al.*, 1987). Since this reality is external to the observer, objectivity can be maintained in observing, recording and deducing results from those observations. Quantitative measures should be used to remove the scope for interpretative distortion of reality. For the axiomatic realist, the fundamental limitation with research involving not only humans but living systems in general, is the lack of repeatability and lack of control over the variables (Kirk and Miller, 1991).

For the purposes of this thesis and the papers featured, a realist perspective is adopted that recognises that a psychological reality can be objectively observed through careful methods design, whilst recognising that the interpretation of those observations are filtered through the researcher's perspectives (Pawson and Tilley, 1997).

2.1.2 Epistemology

Following on from the ontology of the research, consideration of epistemology is required; that is the nature of knowledge and the relationship between the researcher and the research domain (Creswell, 1994). To maintain philosophical integrity there should be a clear route from ontological to epistemological assumptions. Adopting an existential ontology leads one towards a critical theory of knowledge generation along the lines of Habermas (1991, 1986) where the researcher is an integral part of the research domain.

Quantitative or axiomatic research requires an objective researcher that maintains a distance from the research domain so as to maintain the purity of the data gathered. There should be a clear distinction between the researcher, the research domain and the grounds upon which the knowledge is formulated.

2.1.3 Axiology

Axiology considers the role of values and the extent to which rules can be extrapolated from the knowledge gained about our reality (Creswell, 1994). If the researcher is objectively detached from the research domain (coming from a

positivist perspective), as in quantitative research, it is assumed that data will be value-free and bias in raw data will be removed through careful experiment design. However, if the research is from a realist perspective, the research is value laden. The researcher is biased by world views, cultural experiences and upbringings. These affect research findings, but the methods are pragmatically chosen to answer the question. These can be either qualitative, quantitative or mixed methods studies.

2.1.4 Rhetoric

The use of language within research changes as one moves along the ontological scale. Quantitative research tends to adopt a formal and impersonal language, developing definitions and equations upon which value-free data can be related (Creswell, 1994). Qualitative research uses informal language and story-telling is frequently found to develop arguments which explain the value-laden data.

2.1.5 Methodology

Finally, there is the methodology that is adopted for conducting research, which should reflect the assumptions concerning ontology, epistemology, axiology and rhetoric (Creswell, 1994). The quantitative use of questionnaires is consistent with the researcher's ontological position, i.e. that observations can be made that are external to the reality of the observer.

2.1.6 Philosophical conclusion

Professionals who work in healthcare often see all the difficulties and hurdles that can prevent research from succeeding or even being attempted. Beyond risk factors identified by statistical analysis there is no substitute for clinical experience (NHS Scotland, 2016). Clinical experience suggests that by understanding and probing the mechanism of change one can understand the impact of an event or an intervention. The systematic nature of healthcare and the positioning of the person within it, combined with real life clinical experience of work with the people who have had a diagnosis of a chronic health care problem and research experiences, have led the researcher to embrace a realist perspective in undertaking research. This represents a philosophy that is aligned through the levels identified by Creswell (1994) and is consistent with the research domains being explored. It also resonates with the triarchic approach of evidence-based practice and the seminal definition of Sackett et al (1996), which suggests that evidence-based practice occurs when external

research evidence is applied with expertise and in the light of patient preferences; others have also emphasised the role of context in framing evidence-based practice (Foster et al, 2013).

2.2 Autoethnography

Autoethnography is a form of qualitative research in which an author uses self-reflection and writing to explore anecdotal and personal experience and connect this autobiographical story to wider cultural, political, and social meanings and understandings. In doing autoethnography, the author confronts "the tension between insider and outsider perspectives, between social practice and social constraints" (Reed-Danahay, 2009). Hence, autoethnography is a research method that:

- Uses a researcher's personal experience to describe and critique cultural beliefs, practices and experiences
- Acknowledges and values a researcher's relationships with others
- Uses deep and careful self-reflection - typically referred to as "reflexivity" - to name and interrogate the intersections between self and society, the particular and the general, the personal and the political
- Shows people in the process of figuring out what to do, how to live, and the meaning of their struggles
- Balances intellectual and methodological rigor, emotion and creativity
- Strives for social justice and to make life better

Adam, Holman Jones, & Ellis (2015)

2.2.1 Researchers auto-ethnographic stance

Given the nature and style of writing in auto-ethnography I will use the first person in this section, and subsequently when my personal reflections and experiences have an impact on the development and progression of my research.

I come from a solid working class family, growing up just outside Cambridge. Education was valued, but with the main aim of getting a job, as soon as was possible. This also came with an implicit understanding that you 'know your place', and that this was more important than 'bettering yourself'. I recall very clearly a

week before I started my undergraduate degree my nana saying that "You should get a job in the co-op and find a husband". I am the first and currently only member of my family to have A-levels. The wider family thought that that was more than enough academic freedom and could not understand what attraction University could possibly hold. To be honest, I wasn't sure either, but I knew that it was something that I wanted to try. Something that was the obvious next step!

Perhaps my view of university and my subsequent research career has skipped along with the same thought in mind... well that is the obvious next step! Looking back, it feels like I was brought up project managing my life, fitting everything in, delivering on time and on target. My primary focus has always on being able to 'do' things. That has led me to feel most happy whilst undertaking tasks and adapting pragmatically to problems. The problem is that most of research isn't actively doing stuff, a lot of it is sitting and thinking, and reading and writing. I hate writing! So how did I end up here?

I love having a question and then working it through with patients, carers, spouses, Allied Health Professionals, medical doctors, nurses, scientists, and researchers. I love the excitement of unpicking what that answer actually means to all the stakeholders. I love understanding how it informs and impacts people and their choices.

Every person I have worked with has given me at least their time. A vast majority have given me encouragement (often in combination with coffee and criticism). With some I have shared tears and laughter. One with some of his last breaths reinforced how his experiences can live on in my research. I got here to thank all those people and to try to represent their voices in what happens to people within the healthcare environments (practitioners or patients) now and in the future.

2.3 Summary

I have adopted a realist perspective in undertaking the research included in this thesis. This supports the inclusion in the research process of the perspectives of both the researcher and the participant/patient.

In the following chapters I will review the methods used and how these affect the interpretation of the data within the publication and make some comments about its usefulness to the practice.

Chapter 3 Single Time Point Studies

This chapter includes papers reporting primary research investigating the prevalence and presence of a psychological phenomenon. They provide examples of the first element of an investigation where the researcher seeks to identify if the phenomenon exists or is present within the study population.

Central to the development of my research has been the application of health psychology to better understand clinical practice. These two papers demonstrate this. The first explores the occurrence of Post-Traumatic Stress Disorder (PTSD) in a population with Chronic Obstructive Pulmonary Disorder (COPD). The second investigates the presence of positive psychological change (PPC) following treatment for head and neck cancer (HNC) within one year of treatment.

3.1 Paper 1 - Clinical experience leading to the research work and production of the paper

This research came about through the interest of one of the co-authors (Rupert Jones). His interest in PTSD arose from clinical practice as a general practitioner, when he found that patients with PTSD presented a difficult clinical challenge. They had a marked reluctance to admit their problems, and would go to some lengths to find alternative explanations for their symptoms, often ashamed to admit they had the symptoms of PTSD. Typical presenting features included somatic symptoms, negative cognition and incongruous behaviour. Patients with PTSD struggled with relationships and made comments that Rupert interpreted to mean that they felt alone in the world with their problems. Eventually, when they came to accept the diagnosis, there was a huge improvement in their demeanour and their relationships with their spouses and families.

Rupert received funding from the Royal College of Physicians to undertake a project to identify PTSD in chronic cardiopulmonary populations, which included Ischaemic Heart Disease and COPD. I applied to be the research assistant on the COPD element of the research. The project interested me as I had been working with patients with various medical conditions and treatment side effects. My official role with patients to this point had been to help with management of their physical issues, but I spoke with them informally about psychological challenges. They had spoken to me about the struggle of dealing with diagnosis, feared recurrence of their original illness and

how living day to day was shaped by their experiences.

Acute medical conditions, such as myocardial infarction, may trigger PTSD, (Tedstone & Tarrier, 2003) but less is known of the potential for chronic medical conditions to cause PTSD (Alonzo, 2000). COPD is a common, progressive debilitating condition which causes dyspnoea, coughing and often severe anxiety (Karajgi et al., 1990; Wagena et al., 2005). People with COPD may also experience acute exacerbations which can cause life threatening breathlessness. COPD causes 30,000 deaths per year in the UK (Office for National Statistics, 2000), with many deaths occurring during an exacerbation. Alonzo (2000) hypothesised that COPD may be a precipitant of PTSD, but commented that there were no published data to confirm or refute the hypothesis. The interaction between COPD and PTSD may cause clinically important problems via various mechanisms. These mechanisms include effects mediated through classical symptoms of hyperarousal, avoidance and intrusive thoughts and also through health related behaviour such as smoking, exercise and diet.

3.1.1 Research Questions

The work was developed to answer three questions:

- 1) What is the prevalence of PTSD in patients with COPD referred to Pulmonary Rehabilitation (PR)?
- 2) Do PTSD symptom scores fall following PR?
- 3) What is the relationship between PTSD symptom scores and changes in exercise tolerance and health status measures in patients with COPD?

3.1.2 Study Design

We chose to examine a population of patients who were attending PR. This was a convenient sample; they were already being selected and assessed for PR. This approach also offered the possibility of observing changes in outcomes in those with and without PTSD after PR.

In stage one it was decided to undertake an assessment of the prevalence of PTSD in COPD patients referred to PR without a control group. If it was found that the prevalence of PTSD was at a clinically important level, a more detailed study would be conducted as stage two. We did not reach the threshold for stage two.

3.1.2.1 Defining Posttraumatic Stress Disorder

In the psychometric literature at the time of conducting the research disseminated in paper 1, trauma was defined as an event that involved "actual or threatened death, serious injury, or other threat to one's physical integrity" (APA, 2000, p. 463).

Posttraumatic stress disorder (PTSD) is a common psychological and physiological response to a traumatic event. According to the DSM-IV-TR (Diagnostic Statistical Manual; APA, 2000), the essential feature of this disorder is that symptoms occur as a direct result of exposure to a traumatic event involving either direct personal experience, or witnessing or learning about an event that involves another person (APA, 2000). The person must also feel "intense fear, helplessness, or horror" (APA, 2000, p. 463) in response to the event, as well as persistently re-experiencing the of traumatic event, avoiding trauma-related stimuli, demonstrating a numbing of general responsiveness, and experiencing increased arousal and significant distress or impairment for at least one month after trauma (APA, 2000). Subsequent to the data collection, the Diagnostic Statistical Manual was revised, with a reduction in the severity of the trauma and an increase in the range of symptoms being included. However, no revisions were made to the questionnaires used within research to measure PTSD.

3.1.3 The prevalence of PTSD and changes in PTSD symptoms following pulmonary rehabilitation.

The Prevalence of Posttraumatic Stress Disorder in Patients Undergoing Pulmonary Rehabilitation and Changes in PTSD Symptoms Following Rehabilitation

Rupert C. M. Jones, MD, Sam A. Harding, MSc, Man Cheung Chung, PhD, and John Campbell, MD

- **PURPOSE:** Posttraumatic stress disorder (PTSD) is a common serious condition, which, although treatable, is often undetected. We investigated the prevalence of PTSD in patients with chronic obstructive pulmonary disease (COPD) referred to pulmonary rehabilitation and the impact of rehabilitation on PTSD symptoms.
- **METHODS:** Patients with COPD attending pulmonary rehabilitation programs in South West England completed cross-sectional and longitudinal surveys. Outcome measures included the Posttraumatic Diagnostic Scale, Impact of Events scale, Incremental Shuttle Walking Test, Medical Outcomes Short Form 12, Hospital Anxiety and Depression scale (HADS), and Chronic Respiratory Questionnaire. Questionnaires were completed at face-to-face interviews with participants 1 week before commencing pulmonary rehabilitation and at the end of the program.
- **RESULTS:** Patients ($N = 100$), mean age 68 years, 65% men, served as subjects. Seventy-four participants reported traumatic experiences (37 related to lung disease) and 70 completed the pulmonary rehabilitation program. Eight of 100 participants met diagnostic criteria for PTSD. Participants with PTSD reported worse health status than those without PTSD. After pulmonary rehabilitation, exercise capacity and quality of life scores improved significantly, but PTSD symptom severity did not change.
- **CONCLUSIONS:** PTSD was present in 8% of COPD patients referred for pulmonary rehabilitation. After rehabilitation, participants with PTSD improved more in respect to anxiety and disease-specific health status than those without PTSD. PTSD symptoms did not improve following rehabilitation, despite its positive effects on HADS scores, exercise, and health status in this cohort.

KEY WORDS

chronic obstructive pulmonary disease
posttraumatic stress disorder
pulmonary rehabilitation
quality of life

Author Affiliations: Respiratory Research Unit, Peninsula Medical School, Plymouth (Dr Jones and Mr Harding) and Clinical Psychology Teaching Unit, University of Plymouth, Peninsula Allied Health Centre, Plymouth (Dr Chung), and General Practice and Primary Care, St Lukes Campus, University of Exeter, Exeter (Dr Campbell), England, UK.

Corresponding Author: Rupert C. M. Jones, MD, Respiratory Research Unit, Peninsula Medical School, N21 11TC Bldg, 1 Davy Rd, Plymouth PL6 8BX, England, UK (rupert.jones@pms.ac.uk).

Chronic obstructive pulmonary disease (COPD) is a common, progressive, and debilitating condition that causes dyspnea, cough, and disability.¹ Progressive breathlessness is frequently associated with anxiety, panic fear disorder, and depression.²⁻⁴ People with COPD may also experience acute exacerbations that can cause life-threatening breathlessness.¹ Functional decline in COPD is associated with reduced physical

activity and physical deconditioning.⁵ People with COPD frequently become socially isolated and may be blamed for their own disease because they smoked.⁶

Posttraumatic stress disorder (PTSD) is a common serious condition, which, although treatable, is often undetected. In PTSD, persistent symptoms of hyperarousal, avoidance, and reexperiencing of the event

are triggered by reminders of a traumatic experience.⁷ Acute medical conditions, such as myocardial infarction, may trigger PTSD,⁸ but less is known of the potential for chronic medical conditions to cause PTSD.⁹ Alonzo⁹ hypothesized that COPD may be a precipitant of PTSD, but there are no published data to confirm or refute the hypothesis.

If people with COPD develop PTSD triggered by COPD symptoms, hyperarousal has the potential to aggravate breathlessness. The sensations of breathlessness and anxiety are closely related and anxiety caused by dyspnea is a normal response to not being able to breathe.¹⁰ In some people with COPD, a vicious circle can develop in which breathlessness can cause a disproportionately high level of anxiety and panic, which in turn heighten the perception of severe breathlessness.^{4,10}

Avoidance behavior associated with PTSD has the potential to impair physical and social function. People with COPD avoid physical activity compared with nonaffected individuals and thereby reduce the symptoms of breathlessness.¹¹ Physical inactivity leads to progressive functional and social limitations.⁵ Avoidance in PTSD, as defined by the *Diagnostic and Statistical Manual of Mental Disorders* (4th ed) (*DSM-IV*), includes a "feeling of detachment or estrangement from others; restricted range of affect and a sense of foreshortened future"^{7(p463)}; thus, PTSD triggered by COPD symptoms may aggravate impaired quality of life and social function. Furthermore, PTSD has been shown to be associated with higher levels of cigarette smoking among affected individuals,¹² and smoking is a major cause of deterioration in people with COPD.¹³

Pulmonary rehabilitation is an effective therapy for COPD.⁵ Pulmonary rehabilitation involves exercise training, education, and psychosocial support delivered by a multidisciplinary team for groups of patients with chronic lung disease. The exercise program provides graduated exposure to breathlessness and as such is similar to exposure techniques used in cognitive-behavioral therapy, including trauma-focused behavior therapy for PTSD.¹⁴ In common with treatments for PTSD, pulmonary rehabilitation involves allowing patients to share their experiences and reactions to traumatic situations and includes counseling in groups, expressing hidden fears, guilt, anger, and denial. In rehabilitation, relaxation and breathing exercises to reduce anxiety associated with dyspnea are taught.¹⁴ Although pulmonary rehabilitation may be expected to help patients with COPD who have a high level of anxiety and impaired social function, it is not known if pulmonary rehabilitation will improve PTSD symptoms.

The purpose of this study was to examine (1) the prevalence of current PTSD in patients with COPD referred to pulmonary rehabilitation, (2) any changes

in PTSD symptom scores following pulmonary rehabilitation, and (3) the relationship between PTSD symptom scores and changes in exercise tolerance and health status measures in patients with COPD.

METHODS

The study was approved by the South West local research ethics committee. Participants were recruited from patients who had been invited to take part in pulmonary rehabilitation at various programs in Devon, in South West England. All patients referred to pulmonary rehabilitation were initially assessed for their suitability to take part in pulmonary rehabilitation and, if selected, were subsequently assessed for inclusion in the study. Figure 1 shows the different stages of assessment and progression through pulmonary rehabilitation and the study.

Members of the pulmonary rehabilitation team checked that patients were willing and suitable to take part in the rehabilitation program. The rehabilitation assessments were 2-fold; clinical

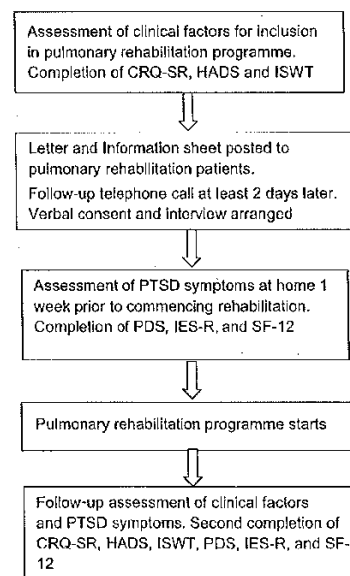


Figure 1. Flowchart showing different stages of assessment. Abbreviations: CRQ-SR, Chronic Respiratory Questionnaire-Self-Report; HADS, Hospital Anxiety and Depression Scale; IES-R, Impact of Events Scale-Revised; ISWT, Incremental Shuttle Walking Test; PDS, Posttraumatic Stress Diagnostic Scale; PTSD, posttraumatic stress disorder; and SF-12, Medical Outcomes Short Form 12.

assessment to ensure suitability and COPD status assessed by exercise tests and questionnaires including the Incremental Shuttle Walking Test (ISWT),¹⁵ which is an externally paced test of maximal exercise capacity; the Hospital Anxiety and Depression Scale (HADS),¹⁶ a self-complete 14-item scale with 2 domains (anxiety and depression); and the self-reported version of the Chronic Respiratory Questionnaire (CRQ SR),¹⁷ which is a disease-specific health status measure with 4 domains (dyspnea, fatigue, emotional function, and mastery).

Only patients with a physician diagnosis of COPD confirmed by spirometry who had been accepted for the pulmonary rehabilitation program were invited to take part in the study. Exclusion criteria were current treatment of major physical comorbidities, major psychiatric illness, confusion, learning disability, or other conditions impairing ability to give informed consent.

Participants were recruited from 3 pulmonary rehabilitation programs: 2 were conducted in the community and 1 in a hospital setting. The programs were similar in terms of their components of exercise, education, and psychosocial support, but 2 programs involved twice-weekly sessions and one of the community-based programs was performed once weekly.

Study Assessments

Those agreeing to take part in the study were interviewed in their own homes at least 1 week prior to start of the pulmonary rehabilitation program. The assessment was conducted prior to the program to avoid asking questions, which might cause distress to patients at the point when they started the program. Participants were invited to complete the Posttraumatic Stress Diagnostic Scale (PDS),¹⁸ the Impact of Events Scale-Revised (IES-R),¹⁹ and the Medical Outcomes Short Form 12 (SF-12).²⁰

The PDS may be used to assess PTSD according to *DSM-IV* criteria. The first part of the PDS consists of 13 questions, which focus on a range of previous traumatic events throughout life that participants may have experienced. The second part (8 questions) assesses PTSD symptoms. If participants had more than 1 traumatic event, they were asked to identify the one that "bothered them the most" and complete the questionnaire accordingly. In the final 20 questions, participants are asked to rate the severity of symptoms according to the rating scale: 0 = not at all; 1 = once a week or less/once in a while; 2 = 2 to 4 times a week/half the time; 3 = 5 or more times a week/almost always. This scale has shown good reliability and validity and good agreement with the Structured Clinical Interview for Diagnosis of PTSD.²¹ The PDS may be used to assess the 6 diagnostic criteria speci-

fied in the *DSM-IV*; the nature of the trauma, the 3 symptom clusters (reexperiencing, avoidance, and hyperarousal), duration of symptoms, and impaired functioning. For example, some respondents may be classified as meeting criteria for reexperiencing symptoms, but not other PTSD symptoms. Only if all 6 criteria are met, is the diagnosis of PTSD confirmed. The PDS may also be scored to produce a quantitative measure of symptom severity for the 3 symptom clusters and the total PTSD score. These are known as symptom severity scores.

The IES-R is a self-reported questionnaire that can be anchored to any specified life event. The scale has 22 items with responses reported using a 0 to 4 Likert scale. The questionnaire focuses on a single episode of trauma, and the respondents are asked to rate how distressing they have found the event in the last 7 days. The original IES assessed only intrusion and avoidance; the revised version added a domain of hyperarousal without changing the existing domains.²² The IES-R may be used to give a score to the impact of an event in terms of intrusive thoughts, avoidance, and arousal, as well as a total score. Both the IES and the IES-R have been widely used and have excellent psychometric properties.^{14,23}

Participants were also invited to complete the Medical Outcomes SF-12.²⁰ The SF-12 is a 12-item, self-administered questionnaire, which is used to assess physical and mental symptoms, social functioning, and quality of life. Each question has between 3 and 6 response options using a Likert-type scale. Scores may be derived for 2 subscales: the physical component summary (PCS) and mental component summary (MCS).

The PTSD questionnaires, SF-12, and all of the pulmonary rehabilitation assessments were completed a second time at the final session of the pulmonary rehabilitation program, 7 to 9 weeks after starting the program. Those who did not complete the program and the postpulmonary rehabilitation assessments were considered to have dropped out. Their data recorded prior to pulmonary rehabilitation was used for assessing the prevalence of PTSD and for cross-sectional analysis.

Statistical Analysis

Data were collated and analyzed using SPSS® (version 14). Descriptive statistics were undertaken to define the participant characteristics and outcome measures. For normally distributed data, comparing means was undertaken using the *t* test (independent samples or paired samples as appropriate). Where data were not normally distributed or failed to meet other assumptions of the *t* test, nonparametric tests were employed. Correlations were undertaken using the Pearson

correlation coefficient as the data were approximately normally distributed. Linear regression analysis was performed to examine whether PTSD symptoms affected outcomes following pulmonary rehabilitation, for example, the scores for the CRQ-SR, the IADS, and the ISWT. For each outcome variable, the final outcome score was entered as the dependent variable, and the initial outcome score and the initial PTSD symptom score were entered as independent variables.

Some missing data were encountered especially in relation to pulmonary rehabilitation records including the baseline characteristics, spirometry, and pulmonary rehabilitation outcome measures. Despite endeavors to locate missing data from the rehabilitation teams or from alternative sources, such as hospital or primary care records, some data could not be obtained.

RESULTS

Of 146 participants attending the pulmonary rehabilitation program, 122 (83.6%) met the inclusion criteria and were invited to take part in the study. One hundred (82%) were willing and available to participate in the study, their mean age was 68 years (SD = 8.2), and 65 (65%) were men.

Spirometry data were available on 85 of the study participants: airflow obstruction was classified according to GOLD guidelines as GOLD II in 27/85 (32%); GOLD III in 41/85 (48%); and GOLD IV in 17/85 (20%).¹ Sixteen were current smokers, 77 were ex-smokers, and only 7 had never smoked. For all participants, the mean total cigarette consumption, expressed in pack years, was 45 (SD 28, range 0–140). One pack year is 20 cigarettes per day for 1 year.

Posttraumatic Stress Disorder

Using the checklist of traumatic events in the PDS, traumatic events were reported by 73 of 100 participants. Many participants reported more than 1 event, and between them, the 73 participants reported a total of 192 traumatic experiences. Twenty-seven participants reported no traumatic experiences. Thirty-seven participants reported traumatic experiences related to their lung disease. When asked to select which traumatic experience "bothered them the most," 24 participants reported that their most traumatic experience was related to their lung disease (Table 1). Traumatic events related to their COPD were mostly caused by acute exacerbations. COPD specific causes were severe breathlessness often accompanied by panic (11/24, 46%), hospitalization (7/24, 29%), pneumonia accounted for (2/24, 8%), pneumothorax (2/24, 8%), and living with COPD (2/24, 8%).

Table 1 • FREQUENCY AND NATURE OF THE MOST TRAUMATIC EVENTS THAT PARTICIPANTS REPORTED

Most traumatic event (n = 73)	n (%)
Respiratory	24 (33)
Other illness	11 (15)
Bereavement	14 (19)
Illness in loved one	10 (14)
Accident/war	10 (14)
Relationship/social problems	4 (5)

Prevalence of PTSD

Eight of 100 participants met the PDS criteria for PTSD. Six of the 8 participants with PTSD reported a traumatic event related to their lung disease. The number of participants meeting the criteria for re-experiencing was 38/100, for avoidance 15/100, and for hyperarousal 13/100. None of the participants were aware of a prior diagnosis of PTSD.

PTSD and health status

PTSD was associated with significantly higher levels of anxiety and CRQ-SR total and emotion domain scores (but not fatigue, dyspnea, or mastery) than those without PTSD (Table 2). Furthermore, the MCS of the SF-12 (but not PCS) was lower in participants having PTSD compared with those not having PTSD (lower scores on the SF-12 indicate worse health status). The ISWT distance achieved did not differ significantly between those with or without PTSD. Although the numbers were small with only 8 participants with PTSD, the assumptions of the *t* test were met; the analysis was repeated using the Mann-Whitney *U* test and the same results were found. There was no correlation between total PDS symptom severity and exercise tolerance (ISWT distance, $r = -0.39$, $n = 84$, $P = .72$) or between total PDS symptom severity and dyspnea (CRQ-SR dyspnea domain, $r = -0.16$, $n = 88$, $P = .13$).

Smoking and PTSD status

No significant associations between smoking status (cigarette consumption or pack years) and measures of PTSD severity (PDS symptom severity score or IES-R total score) were found. Current smokers, ex-smokers, and never smokers were similar with respect to PTSD total symptom score (Kruskal-Wallis) and in the proportion of these participants identified as having PTSD in this study (chi-square).

Table 2 • THE DIFFERENCES IN HEALTH STATUS (MEAN $\pm$ SD) AND EXERCISE LIMITATION BETWEEN PARTICIPANTS WITH OR WITHOUT PTSD^a

	PTSD (n = 8)	No PTSD (n = 92)	p ^b
CRQ-SR total	3.78 $\pm$ 0.97	2.99 $\pm$ 0.60	.026
CRQ-SR emotion	4.27 $\pm$ 1.32	3.25 $\pm$ 0.86	.038
CRQ-SR fatigue	3.82 $\pm$ 1.39	2.88 $\pm$ 0.60	.061
CRQ-SR dyspnea	2.53 $\pm$ 1.06	2.08 $\pm$ 0.48	.235
CRQ-SR mastery	4.45 $\pm$ 1.34	3.77 $\pm$ 1.23	.171
SF-12 PCS	29.90 $\pm$ 6.61	32.64 $\pm$ 8.16	.514
SF-12 MCS	25.98 $\pm$ 6.84	32.65 $\pm$ 7.65	.009
HADS anxiety	11.88 $\pm$ 4.09	7.33 $\pm$ 4.16	.010
HADS depression	8.13 $\pm$ 2.80	6.37 $\pm$ 3.29	.106
ISWT (m)	163.75 $\pm$ 87.66	189.88 $\pm$ 114.55	.533

Abbreviations: CRQ-SR, Chronic Respiratory Questionnaire—Self-Report; HADS, Hospital Anxiety and Depression Scale; ISWT, Incremental Shuttle Walking Test; MCS, Mental Component Score; PCS, Physical Component Score; PTSD, posttraumatic stress disorder; SF-12, Medical Outcomes Short Form 12.

^aFor all tests, higher scores indicate worse health status, except SF-12 and ISWT, where lower scores indicate worse health status.

^bt test.

Changes in PTSD measures after pulmonary rehabilitation

Participants completing the rehabilitation program ($n = 70$) were similar to those who did not complete the program ($n = 30$) with respect to age, gender, airflow obstruction (% of predicted forced expiratory volume in 1 second), smoking status and pack years, ISWT, SF-12 PCS, PDS, and IES-R scores. After pulmonary rehabilitation, exercise capacity and all scores derived from CRQ-SR, SF-12 MCS, and HAD scales improved significantly in this cohort of participants (Table 3). However, PTSD symptom severity measured by PDS or IES-R did not change significantly:

mean PDS symptoms severity score, 5.0 (8.9) before, 6.1 (7.7) after, $P = .52$; mean IES-R total, 4.6 (10.8) before, 5.1 (12.2) after, $P = .24$. Furthermore, there were no significant changes in the domain scores of the PDS and IES-R after the program.

PTSD symptom scores, exercise tolerance, and health status

Linear regression analyses failed to show any significant differences in outcomes between the PTSD and non-PTSD participants for the rehabilitation outcome variables, namely the domain and total scores for the CRQ-SR, the 2 subscales of the HADS, and the ISWT

Table 3 • CHANGES IN OUTCOMES FOLLOWING PULMONARY REHABILITATION IN ALL PARTICIPANTS (MEAN $\pm$ SD)^a

Measure	Before pulmonary rehabilitation	After pulmonary rehabilitation	p ^b
CRQ-SR total score	3.71 $\pm$ 0.97	4.50 $\pm$ 0.94	.001
CRQ-SR dyspnea	2.49 $\pm$ 1.03	3.12 $\pm$ 1.14	.001
CRQ-SR fatigue	3.73 $\pm$ 1.37	4.54 $\pm$ 1.62	.001
CRQ-SR emotion	4.18 $\pm$ 1.32	5.02 $\pm$ 1.35	.001
CRQ-SR mastery	4.39 $\pm$ 1.33	5.29 $\pm$ 1.19	.001
ISWT (m)	191 $\pm$ 105	246 $\pm$ 130	.001
SF-12 PCS	33.3 $\pm$ 8.3	33.0 $\pm$ 7.4	.735
SF-12 MCS	32.2 $\pm$ 7.8	35.5 $\pm$ 7.5	.001
HADS anxiety	7.7 $\pm$ 4.0	5.4 $\pm$ 3.9	.001
HADS depression	6.4 $\pm$ 3.2	5.4 $\pm$ 3.8	.025
PDS symptoms severity	5.0 $\pm$ 7.6	6.2 $\pm$ 7.7	.239
IES-R total	4.5 $\pm$ 10.8	5.1 $\pm$ 12.2	.520

Abbreviations: CRQ-SR, Chronic Respiratory Questionnaire—Self-Report; IES-R, Impact of Events Scale-Revised; HADS, Hospital Anxiety and Depression Scale; ISWT, Incremental Shuttle Walking Test; MCS, Mental Component Score; PCS, Physical Component Score; PDS, Posttraumatic Stress Diagnostic Scale; SF-12, Medical Outcomes Short Form 12.

^aFor all tests, higher scores indicate worse health except SF-12 and shuttle walking test, where lower scores indicate worse health status.

^bt test.

Table 4 • SUMMARY OF EIGHT LINEAR REGRESSION ANALYSES TO ASSESS THE CONTRIBUTION OF THE INITIAL PTSD SYMPTOM SEVERITY SCORE TO FINAL OUTCOME AFTER CONTROLLING FOR INITIAL OUTCOME SCORE

Outcome variable	n	Standardized coefficients (β) for initial PTSD symptom score	t	P
CRQ-SR				
Total	68	.046	0.45	.66
Dyspnea	68	.046	0.45	.66
Fatigue	70	.008	0.08	.94
Emotion	70	.163	1.62	.11
Mastery	70	.141	1.31	.20
HADS				
Anxiety	70	.049	0.47	.64
Depression	70	.098	0.94	.37
ISWT	69	-.035	-0.52	.61

Abbreviations: CRQ-SR, Chronic Respiratory Questionnaire—Self-Report; HADS, Hospital Anxiety and Depression Scale; ISWT, Incremental Shuttle Walk Test; PTSD, posttraumatic stress disorder.

(Table 4). Neither the initial PTSD symptom score nor its interaction with the initial CRQ-SR total score is a predictor of outcome as represented by the CRQ-SR total score. The analyses indicate that PTSD status does not predict the success of pulmonary rehabilitation.

DISCUSSION

In this study, we investigated the prevalence of PTSD in patients attending pulmonary rehabilitation, the changes in PTSD symptom status following pulmonary rehabilitation, and whether PTSD symptoms predicted changes on exercise tolerance and health status measures. We found that PTSD was present in a minority (8%) of COPD patients referred for pulmonary rehabilitation. The prevalence of PTSD in normal populations has been estimated at between 1% and 8%, the highest figure deriving from a nationally representative sample of younger adults (≤ 54 years old) in the United States.²⁴ In a study of older people aged 55 to 90 years, the prevalence was 1%.²⁵

Higher prevalence rates of PTSD have been reported in people suffering from serious medical conditions.⁸ Using similar diagnostic measures, we have previously reported that 32% of patients with a previous myocardial infarction met the diagnostic criteria for PTSD.²⁶ The prevalence after being given the diagnosis of HIV was 30% in 1 study.²⁷ The prevalence of PTSD in people with COPD was in line with the findings of a study of psychological diagnoses in

patients hospitalized with COPD in which only 1 patient in 50 had PTSD.²⁸

Of the 8 participants with PTSD, 6 experienced traumatic events related to their lung disease. Thus, although the prevalence rate of PTSD in this sample is not high, compared with other traumatic events, respiratory-related traumas were represented as the most frequent factor causing PTSD. A further study including a control group that did not have COPD would be needed to elucidate this issue further.

This study confirmed that those with PTSD reported not only a worse quality of life as measured by HADS anxiety and the MCS of the SF-12 but also worse disease-specific quality of life as measured by the CRQ-SR total and emotion domains. No significant differences were found between those with PTSD and those who did not have PTSD with respect to the HADS depression scores, the PCS of the SF-12, and the dyspnea, fatigue, and mastery scores of the CRQ-SR. These findings are consistent with the assertion that PTSD is primarily an anxiety disorder.⁷

PTSD has been associated with adverse behaviors likely to affect outcomes in COPD such as smoking.²⁹ Smoking is the major cause of the development and progression of COPD¹³ and smoking cessation is a critical component of COPD management.¹ In this study, no evidence was found that PTSD affected smoking status, including current smoking status, daily cigarette consumption, or lifelong consumption of cigarettes.

In the introduction, the suggestion was made that hyperarousal in PTSD may be associated with worsening of breathlessness. There was no evidence in this study to support that hypothesis. A further hypothesis

was that avoidance associated with PTSD would lead to reduced exercise tolerance. There was no evidence that PTSD generally or avoidance specifically was associated with higher levels of perceived breathlessness as measured by the CRQ-SR dyspnea domain or the maximum exercise tolerance as measured by the ISWT. Thus, PTSD has not been shown to affect the exercise tolerance of patients with COPD in this study. The study is relatively small with only 8 participants with a diagnosis of PTSD, and these findings should be considered as preliminary rather than definitive.

We have hypothesized that pulmonary rehabilitation may have a beneficial effect on participant PTSD symptoms. We found that those with PTSD improved more in respect to anxiety and disease-specific health status than those without PTSD. Pulmonary rehabilitation had a substantial beneficial effect on COPD measures including the quality of life and exercise capacity with mean improvement above the minimum clinically important differences (for CRQ-SR domains 0.5 points and for the ISWT 48 meters). However, while disease-specific outcome measures and the anxiety domain of the HAD scale showed substantial improvements, PTSD symptoms did not change, indicating that PTSD symptoms were resistant to the general measures provided by pulmonary rehabilitation. One conclusion from this is that specific treatments, which address the PTSD *per se*, such as trauma-focused cognitive-behavioral therapy, may be needed to improve the PTSD status of people with both PTSD and COPD.

PTSD is present in a small but important proportion of individuals with COPD referred for pulmonary rehabilitation. Participating in such programs may result in improvements in anxiety and respiratory symptoms, especially among those individuals with PTSD. Providers of such programs should be alert to the psychological status and needs of those attending. For those with PTSD, specific treatments may be needed over and above the therapeutic modalities provided by pulmonary rehabilitation.

—Acknowledgments—

We thank East Devon Respiratory Research Group, Gill Ackers and all her colleagues from the Honiton Pulmonary Rehabilitation group, Jane Trott from the Royal Devon and Exeter Foundation Hospital, Judith Wells from the Plymouth Trust Pulmonary Rehabilitation program and Chest Clinic, and Dr Bryantie Shackell from the respiratory research group, Peninsula Medical School.

References

1. Global Initiative for Chronic Obstructive Lung Disease. Global strategy for the diagnosis, management and prevention of

- COPD. 2006. <http://www.goldcopd.com/Guidelineitem.asp?l1=2&l2=1&intId=989>. Accessed September 8, 2008.
2. van Manen JG, Bindels PJ, Dekker FW, IJzermans CJ, van der Zee JS, Schade E. Risk of depression in patients with chronic obstructive pulmonary disease and its determinants. *Thorax*. 2002;57:412–416.
3. Wagena EJ, Arrindell WA, Wouters EF, van Schayck CP. Are patients with COPD psychologically distressed? *Eur Respir J*. 2005;26:242–248.
4. Rose C, Wallace L, Dickson R, et al. The most effective psychologically-based treatments to reduce anxiety and panic in patients with chronic obstructive pulmonary disease (COPD): a systematic review. *Patient Educ Couns*. 2002;47:311–318.
5. Nici L, Donner C, Wouters E, et al. American Thoracic Society/European Respiratory Society statement on pulmonary rehabilitation. *Am J Respir Crit Care Med*. 2006;173:1390–1413.
6. Morgan MDL, Threlfall R, Sanders D. Living with COPD—results of the British Lung Foundation survey. *Eur Respir J*. 2001;18(suppl 33):488.
7. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders*. 4th ed. Washington, DC: American Psychiatric Association; 1995.
8. Tedstone JE, Tanier N. Posttraumatic stress disorder following medical illness and treatment. *Clin Psychol Rev*. 2003;23:409–448.
9. Alonzo AA. The experience of chronic illness and post-traumatic stress disorder: the consequences of cumulative adversity. *Soc Sci Med*. 2000;50:1475–1484.
10. Bailey PH. The dyspnea-anxiety-dyspnea cycle—COPD patients' stories of breathlessness: "It's scary/when you can't breathe." *Qual Health Res*. 2004;14:760–778.
11. Pitta F, Troosters T, Spruit MA, Probst VS, Decramer M, Gosselink R. Characteristics of physical activities in daily life in chronic obstructive pulmonary disease. *Am J Respir Crit Care Med*. 2005;171:972–977.
12. Beckham JC, Kirby AC, Feldman ME, et al. Prevalence and correlates of heavy smoking in Vietnam veterans with chronic posttraumatic stress disorder. *Addict Behav*. 1997;22:637–647.
13. Anthonisen NR, Connett JE, Kiley JP, et al. Effects of smoking intervention and the use of an inhaled anticholinergic bronchodilator on the rate of decline of FEV1. The Lung Health Study. *JAMA*. 1994;272:1497–1505.
14. National Institute for Clinical Excellence. *Post-traumatic Stress Disorder: The Management of PTSD in Adults and Children in Primary and Secondary Care*. London, UK: National Institute for Clinical Excellence; 2005.
15. Singh SJ, Morgan MD, Scott S, Walters DJ, Hardman AE. Development of a shuttle walking test of disability in patients with chronic airways obstruction. *Thorax*. 1992;47:1019–1024.
16. Zigmond AS, Snaith RP. The hospital anxiety and depression scale. *Acta Psychiatr Scand*. 1983;67:361–370.
17. Williams JE, Singh SJ, Sewell L, Guyatt GH, Morgan MD. Development of a Self-Reported Chronic Respiratory Questionnaire (CRQ-SR). *Thorax*. 2001;56:954–959.
18. Foa E. The validation of a self-report measure of posttraumatic stress disorder: the Posttraumatic Diagnostic Scale. *Psychol Assess*. 1997;9:445–451.
19. Weiss DS, Marmar CR. The Impact of Events Scale—revised. In: Wilson J, Keane TM, eds. *Assessing Psychological Trauma and PTSD*. New York: Guilford; 1996:399–411.
20. Ware JE, Keller SD, Kosinski M. A 12-item short-form health survey construction of scales and preliminary tests of reliability and validity. *Med Care*. 1996;34:220–233.
21. Foa EB. *Posttraumatic Stress Diagnostic Scale: Manual*. Minneapolis: National Computer Systems; 1995.
22. Horowitz MJ. Impact of Event Scale: a measure of subjective stress. *Psychosom Med*. 2006;41:209–218.

23. Joseph S. Psychometric evaluation of Horowitz's Impact of Event Scale: a review. *J Trauma Stress*. 2000;13:101-113.
24. Kessler RC, Sonnega A, Bromet E, Hughes M, Nelson CB. Posttraumatic stress disorder in the National Comorbidity Survey. *Arch Gen Psychiatry*. 1995;52:1048-1060.
25. van Zelst WH, de BE, Beekman AT, Deeg DJ, van DR. Prevalence and risk factors of posttraumatic stress disorder in older adults. *Psychother Psychosom*. 2003;72:333-342.
26. Jones RC, Chung MC, Berger Z, Campbell JL. Prevalence of post-traumatic stress disorder in patients with previous myocardial infarction consulting in general practice. *Br J Gen Pract*. 2007;57:808-810.
27. Kelly B, Raphael B, Judd F, et al. Posttraumatic stress disorder in response to HIV infection. *Gen Hosp Psychiatry*. 1998;20:345-352.
28. Yellowlees PM, Alpers JH, Bowden JJ, Bryant GD, Ruffin RE. Psychiatric morbidity in patients with chronic airflow obstruction. *Med J Aust*. 1987;146:305-307.
29. Beckham JC. Smoking and anxiety in combat veterans with chronic posttraumatic stress disorder: a review. *J Psychoactive Drugs*. 1999;31:103-110.

3.1.4 Study limitations

3.1.4.1 Sampling considerations

This study was a preliminary investigation performed with patients referred to PR and, as such, they should not be considered representative of all COPD patients. Pulmonary rehabilitation is recommended for patients with functional disability, so people undergoing PR tend to have moderate to severe COPD.

Patients with comorbidities affecting their mobility were specifically excluded; this was to ensure that COPD was the causal factor for any identified PTSD. Participants were recruited from the South West of England and could be unrepresentative of wider COPD populations, for example, there were very few people from ethnic minorities.

Patients with very severe COPD who are unable to leave the house seldom attend PR (National Collaborating Centre for Chronic Conditions, 2004). Those with PTSD may self-exclude by avoiding PR or declining to take part in the research project.

3.1.4.2 Control group

As a prevalence study there was no control group of subjects without COPD, so the finding of eight percent having PTSD is hard to interpret beyond a low incidence of occurrence in the sample population. Future larger controlled studies would be needed to evaluate the role of PTSD in COPD patients, but the clinical threshold to justify a larger study was not met.

3.1.4.3 Interpreting the findings

Only eight of 100 participants met the diagnostic criteria for PTSD and, only six had PTSD related to their lung disease. The small numbers affect the generalisability of the findings as atypical individuals may skew the results. Statistically there was insufficient power to show significant differences in Quality of Life (QoL) and exercise capacity in those with and without PTSD. For the same reason, changes in PTSD scores following PR must also be considered as preliminary.

I noted that the prevalence of PTSD diagnosis was higher in patients in the early stages of the project and this may have several explanations, one of which is that we used different interviewers. Marked differences in prevalence were noted ranging from 1.3% to 50%. The interviewers were all trained in a similar way, but different

interviewers may have subconsciously influence participants' responses. Patient characteristics like the socioeconomic status and exposure to military service may also affect the prevalence of PTSD. While the numbers were small, there were no apparent explanations for these differences amongst patient characteristics. For example, researcher four saw participants from deprived urban areas to wealthy rural areas and consistently found a low prevalence rate. It might also be that the first set of participants were seen on their own at the PR venue, whereas, I saw all participants in their homes, often with their spouses/carers. It maybe that these people had more social support and therefore developed fewer PTSD symptoms, or that they were less willing to reveal their concerns in front of their spouses.

These differences highlight the critical role of wider context in data collection for psychological research. The same individual may provide different responses in different settings, and researchers may interpret qualitative findings differently given contextual data and their underlying experiences.

3.1.5 Contribution to knowledge and autoethnographic issues

3.1.5.1 Contribution to knowledge

This was the first study to assess the presence and prevalence of PTSD in a COPD population. In this study, the prevalence of current PTSD in 100 patients with COPD referred for PR was assessed. The prevalence was found to be eight percent, which was lower than originally anticipated on the basis of clinical experience. The contribution to knowledge was that a clinical perception (prevalence of PTSD in COPD patients) was evaluated in a rigorous study and found to be exaggerated. There may be many reasons for this over-estimation (which would require a separate study to explore), but the work informed clinicians caring for COPD patients in how the diagnosis may affect those patients.

Appendix 2 has an overview of how the research undertaken in paper 1 was disseminated prior to the paper's publication. It also provides a list of papers where paper 1 has been cited, followed by a quality appraisal of the paper.

3.1.5.2 Autoethnographic issues

I applied for the role of the RA on this project as it appealed academically. I not only felt I matched the personal descriptors, and that I had the skills for the role, but that I would also be able to grow as a researcher. However, at the time I was not sure the

direction that growth would take. In fact I clearly remember being asked 'what training I think I would need if I was offered the job?' As I recall I suggested that I wasn't sure where my areas of deficit for the project lay, so would like to meet with the team once I had become familiar with the project. I cannot recall or find evidence of any formal training undertaken during this project, but there was lots of informal development.

This was the first time I had experience of conducting primary research with individuals in their home environment. The experience of lone working, and following University guidelines on safety and alerting people to my location etc. was new. I appreciate that operating procedures such as lone worker policy are there for the safety of the researcher and the protection of the university, it was however still a challenge to adapt. I felt very uncomfortable attending people's homes, asking them to trust me sufficiently to open up and talk to me about traumatic events in their life, but all the time having processes in place that indicate that they may want to harm me.

I was learning that the duty of care extended as much to the research team (myself in this instance) as to the research participants.

Another challenge was how to deal with people disclosing or revealing significant personal issues that may negatively impact on the participant's health and wellbeing. The issue was that I was interacting with them as a researcher not a clinician. However, they may well see me as having a dual role, and potentially able to influence their care. When I sought consent from potential participants I had to bear in mind my professional role. Although at the time I was not a qualified health psychologist, I was potentially in a perceived position of authority. Potential participants may have been concerned about negative consequences if they refuse to help, or indeed hoped for additional professional interaction that would not fall within a research role. The challenge for participants would be if they could not distinguish between requesting help from me as a researcher, or hoping to gain access to services via me as a perceived clinician. I faced a similar challenge in maintaining clarity over my role. In the context of an open interview, which parts related purely to the research questions, and which could have impact outside my remit and might require onward referral? This also raised potential ethical considerations of required consent to disclose information given within the confidential research setting.

A concrete example of this internal conflict occurred when a participant revealed that she had been sexually abused as a child, then physically abused by her first husband, and how she was now suffering following the death of her second husband to cancer. These life experiences were all revealed in light of our discussions around traumatic events. The challenge arose when she raised her desire to end her own life.

Prior to starting interviews, the team had not formally constructed a policy for safeguarding in this type of situation. So, when the participant revealed her current state, I ended the research interview, explained that I had concerns about leaving her, and asked if she wanted to talk more about her situation. She was open to talking, so we discussed her interaction with her general practitioner, which she perceived as a failing relationship. With permission, I talked to her general practitioner who then attended and supported the patient.

3.1.7 Future research questions

I undertook many of the interviews in participants' own homes, frequently with their spouses, hearing not only the participants' stories, but also how these events impacted those closest to them. This led me to think about how those spouses may develop secondary trauma. Secondary Trauma is a concept developed by trauma specialists Beth Stamm, Charles Figley and others in the early 1990s as they sought to understand why service providers seemed to be exhibiting symptoms similar to PTSD without direct exposure to trauma themselves. This could easily be associated with the experiences of relatives and carers.

There is an unexplored research question around the prevalence of ST and/or PTSD due to COPD in a spouse/carer. This could be expanded in line with the research presented in paper 1, to assess if spousal participation in PR had an additional positive impact on patients' QoL, and other factors included in PR.

3.2 Paper 2 - Clinical experience leading to the research work and production of the paper

Prior to and during my involvement with the research that culminated in paper 1, I was working with people diagnosed with HNC. I started to hear about the struggles patients faced on the day they received their cancer diagnosis, and how they were repeatedly distressed during treatments and when attending clinical appointments. I started to wonder about the potential occurrence of PTSD in this clinical population and began to think of a way to investigate it. Within the hospital clinical team, we discussed at what time points from diagnosis to completion of treatment it would be best to approach patients. We reviewed how the patients could be tracked to ensure the questionnaires were given at the right point. The team felt that a single posting to everyone who met the inclusion criteria would be the most pragmatic way to gain insight into QoL and PTSD in an HNC population. However, following my experiences with the previous research, I felt uncomfortable about sending questionnaires asking about symptoms of PTSD. What if being asked to identify factors relating to PTSD, was in itself a trauma causing distress?

The research undertaken for paper 2 was unfunded, with my time provided on a voluntary basis. Support from the hospital covered only printing and postage costs. This meant that no additional RA time was available for face to face visits, and only a telephone number and hospital email address were given in order for the potential participants to contact me.

I thought that we could still investigate the phenomenon, but using a different more positive lens. The experience of a traumatic or extremely stressful event may be sufficient to challenge a strongly held set of assumptions about the world and the self. In 1975, Parkes used the phrase 'assumptive world' to refer to people's view of reality, defined as *"a strongly held set of assumptions about the world and the self which is confidently maintained and used as a means of recognising, planning and acting.... Assumptions such as these are learned and confirmed by the experience of many years."* (Parkes, 1975. p152). According to this theory, we are rarely aware of the fundamental elements of our assumptive world; the minor disappointments, challenges and failures of day-to-day life seldom bring them to light. It has been said that they are conservative cognitive schemes that resist change and disconfirmation (Janoff-Bulman and Schwartzberg, 1991). The questioning of the basic assumptions

is what fractures the assumptive world and triggers the rebuilding of them to accommodate new realities (Janoff-Bulman and Schwartzberg, 1991). A life-threatening illness such as cancer could be sufficient to shatter a person's assumptive world. In some people, this may lead to the development of PTSD, or the increasingly recognised phenomenon of PPC whereby a person's reactions to the challenge are beneficial to one or more areas of their life.

3.2.1 Research Question

After the completion of treatment for Head and Neck Cancer, what are the demographic, clinical, and psychological factors associated with PPC that occur in the acute period, defined as between 3 and 12 months?

3.2.2 Study Design

In the UK in 2013, 7,591 people were diagnosed with oral cancer, making this the 16th most common cancer diagnosis (Cancer Research UK, 2015). Unlike breast cancer, the population to engage in research at one treatment centre is small. It was therefore important to maximise recruitment in any single site study.

It was specifically decided not to approach people while attending clinic appointments, as the researcher had been informed by people that had received treatment for HNC that, even in routine follow-ups, they felt anxious from the point of receiving the appointment letter until after seeing the consultant. They also did not want to stay in the hospital to complete a survey after their review. The practical solution to this was a postal survey. Undertaking the survey annually reduced the number of times this data had to be centrally requested. It also meant that the hospital notes were less likely to be in use by the clinical team, so requesting them would not hamper the patient's clinical treatment or review. This reduced the burden on hospital resources and the researcher. It was hoped that this method would also minimise the burden on the potential respondents.

I had permission to access the Somerset Cancer Register database at Derriford Hospital (Plymouth). All new cancer diagnoses are required to be entered on to this system as part of a national monitoring programme. The administrator of this database ran a data search within this database and supplied the names, addresses, dates of birth, gender, diagnosis and treatments of people treated by the Head and Neck Directorate in the preceding year.

To see if PPC was present in a HNC population within 1 year of treatment a single time point study was required. To this end, we decided to use a cross-sectional study design. Cross-sectional studies have several characteristics that make them attractive to researchers. They are relatively inexpensive, quick and easy to do, are useful for generating and clarifying hypotheses, piloting new measures or technology and can lay the groundwork for decisions about future follow-up studies (Kraemer, 1994). They provide information about group differences or inter-individual differences (Miller, 2007). They do not, however, provide information about changes or inter-individual differences in intra-individual change (Miller, 2007; Wohlwill, 1973).

3.2.2.1 Psychometric Measures of Positive Psychological Change

In the 1990s, a number of self-report psychometric measures were developed including the SLQ. However, only one study has compared different measures of PPC by looking at the structural classifications of the measures (Joseph *et al.*, 2004). Joseph *et al.* (2004) recruited 176 adults, who had experienced a range of distressing life events. These people completed the Perceived Benefit Scales, the Thriving Scale, and the Posttraumatic Growth Inventory. Confirmatory factor analysis found that all of these sub-scales loaded highly on a single component, which the authors believe is PPC. There was also a suggestion of three second-order components of interpersonal relationships, self-perception, and spirituality. This indicates that many, if not all, developed scales in the field of study may measure an umbrella concept which can be labelled as PPC, but that they may vary in the nature of specific sub-scales.

The SLQ was developed in the same geographical region as the research reported in paper 2 (Sodergren and Hyland, 1998). The mixed disease cohort used to develop the SLQ included a mixed cancer group who were being treated in the same hospital where the wider research project recruited its participants. The SLQ was developed out of a series of interviews that found 18 categories or themes of PPC. These categories included; improved interpersonal relationships, positive influence on others, reappraisal of life, restructuring of life or life style, changes in spirituality, changes in priorities, and seeing illness as a challenge to be overcome. The authors of the scale used language that participants in their interviews had used and refined the phrasing in a similar cohort (Sodergren and Hyland, 1998). Although the interview stage of SLQ development found 18 themes, some of which directly

mapped onto those found in the Post-traumatic Growth Inventory (PTGI), the finalised scale is uni-dimensional.

The SLQ had a 95% completion rate on those surveys that were returned; it was the longest of the measures used. Confirmatory factor analysis of the SLQ by McBride *et al.* (2008) and McBride *et al.* (2009) suggest that a 16 or 24 (respectively) item version of the SLQ with subscales might be valid. The use of the 16 item version would reduce the number of questions being asked by a quarter, which it is hypothesised would have a positive impact on the number of measures returned, as well as increasing the completion rate of the measures that are returned.

The PTGI is currently the most widely used PPC measure in the literature. It has 21 items, so is shorter than the SLQ. However, the PTGI is American and I felt that the framing of the questions, i.e. the language used in the items, was not directly relatable to a UK audience. However, of greater concern are the results of studies attempting to cross-validate the work of Tedeschi and Calhoun (1996). For example, a three-factor structure was discovered in Spanish female immigrants (Weiss & Beiger, 2006), and Chinese cancer patients (Ho, Chan, & Ho, 2004), as opposed to the five subscales found by the scale's developers. The failure to replicate the factor structure of the original PTGI in these samples was attributed to reasons including translational difficulties, that certain items of the PTGI were not applicable to cancer patients, and that the factor structure of posttraumatic growth may vary across different populations (Ho, Chan, & Ho, 2004). Sheikh and Marotta (2005) utilised the original PTGI in a sample of white, middle-aged, cardiovascular disease patients from the United States of America and the United Kingdom and found a two-factor solution, in which the factors were labelled 'general posttraumatic growth' and 'spiritual changes'.

The SLQ was developed to be used with people who have an unspecified disease or illness as a measure of PPC, whereas the PTGI and other measures were developed to assess traumatic events such as earthquakes, rape, and acts of terrorism. As such, the SLQ may be of use to researchers and clinicians who wish to design interventions to target specific areas of PPC and to measure the success or otherwise of psychosocial intervention strategies purported to be of benefit to individuals with chronic illnesses. Given that Linley and Joseph (2004) concluded that there was a need for such measures, it would seem that the use of the SLQ in research was appropriate as it provides baseline data and insight into the natural progression of

PPC development. I concluded that the SLQ has the potential to provide researchers with a multi-dimensional generic tool to measure PPC, and with further investigation into the validity and reliability of a shortened version this could be used effectively in research and clinical settings.

3.2.3 The impact of treatment for head and neck cancer on positive psychological change within a year of completing treatment.

Clinical Paper
Head and Neck Oncology

The impact of treatment for head and neck cancer on positive psychological change within a year of completing treatment

S. Harding¹, T. P. Moss²

¹Department of Health and Social Sciences, University of the West of England, Bristol, UK;
²Centre for Appearance Research, University of the West of England, Bristol, UK

S. Harding, T.P. Moss: *The impact of treatment for head and neck cancer on positive psychological change within a year of completing treatment*. Int. J. Oral Maxillofac. Surg. 2018; 47: 302–308. © 2017 International Association of Oral and Maxillofacial Surgeons. Published by Elsevier Ltd. All rights reserved.

Abstract. Head and neck cancer carries a high level of morbidity and mortality. So why could anyone find having such a disease a positive event? The adversity hypothesis of “what doesn’t kill you makes you stronger” suggests that people can use adversity to develop as human beings. This positive psychological change has received little attention in relation to head and neck cancer. Responses to the Silver Lining Questionnaire, University of Washington Quality of Life Questionnaire, and Short-Form 12 were collected from a postal survey, 3 to 12 months after the completion of treatment for head and neck cancer. Fifty-two (63%) people returned the survey and were included in the analysis. Time since completion of therapy did not show any relationship with positive psychological change. Tumour stage and treatment regimen both had a relationship with positive change. Participants with lower stage tumours had higher levels of positive change than those with tumours of higher stages. Participants who had surgery alone reported more positive change than those who had surgery with radiotherapy. A social factor related to greater change was being married or living with a partner when compared to living alone. Further research would aid the identification of bio-psychosocial factors that influence the development of positive psychological change and inform the development of rehabilitation interventions.

Key words: positive psychological change; post-traumatic growth; head neck cancer.

Accepted for publication
Available online 24 August 2017

Head and neck cancer (HNC) is a relatively uncommon disease with only 6398 new diagnoses in the UK in 2014; however, it carries a high level of morbidity and mortality (50% at 5 years)¹. Factors associated

with this disease have traditionally been studied using a biomedical approach, as the investigators have tended to be clinicians. In the last three decades, psychosocial factors such as health-related

quality of life (HRQoL) outcomes have emerged as an important addition to the conventional clinical outcome measures². Furthermore, in the last 10 years, the phenomenon of positive psychological

change (PPC) following a traumatic experience has sparked the interest of health care professionals working in long-term patient care and interventions.

The construct of PPC has been variously referred to as 'benefit finding' (BF), 'thriving', 'stress-related growth', 'transformational coping', 'post-traumatic growth' (PTG) or 'existential growth', and may concern alterations in the perceptions of oneself, social relationships with family and friends, life priorities, and appreciation of life. The use of these different terms highlights the difficulties in defining processes of growth and conceptualizing the construct. It has been suggested that PTG and BF are distinct constructs that have a conceptual overlap³. However, the scales that have been developed may be argued to measure the same thing differently or in different samples. For example, Scars et al. showed that BF was predicted by personal characteristics (i.e., education, optimism, and hope), but PTG was not⁴. It remains unclear how the two concepts relate to each other, but where BF may start immediately after diagnosis and results from challenges to the individual's cognitive representations, PTG could be hypothesized to develop because of the rumination and restructuring of the self/world relationship that occurs in the weeks, months, and even years following trauma⁵. Because of this temporal and conceptual overlap, it was decided to use PPC as the preferred term in the current study, and it is acknowledged that this study is not differentiating between BF and PTG.

At the time of writing, only five quantitative studies and a systematic review had been published investigating PPC in people who had been treated for HNC⁶⁻¹¹. These investigated the relationship of PPC with various bio-psychosocial factors related to HNC and that have been investigated in HRQoL studies. Harrington et al. assessed the relationship between PPC and treatment regimen, time since treatment, stage of cancer, and diagnosis of further illness, and failed to find any associations⁶. This pattern was reinforced by the findings of Llewellyn et al.⁹ and Hofmaat et al.⁸. Ho et al. found that people with more advanced cancer (stages III and IV) reported lower levels of PPC, but different treatment modalities did not significantly influence PPC⁷. However, Leong et al. failed to find an association between tumour stage and the development of PPC¹⁰. These findings suggest that the biological variables are, at least at present, inconclusive and the impact of demographic factors is equally unclear.

No relationship has been found between gender and PPC^{6,7}, and no published literature has found an impact of age on PPC in HNC, although it has been found that younger participants with breast cancer report higher levels of PPC^{12,13}. Two studies following the treatment for HNC reported a beneficial effect of marriage or stable cohabiting over single status in the reporting of PPC^{7,9}.

Harrington et al. found that in people who have had HNC, dispositional optimism and positive reframing could account for 23% of variance in PPC and that higher levels of religious coping was correlated with greater PPC, but that there was no relationship with anxiety or depression⁶. Once again Llewellyn et al.⁹ supported the findings of Harrington et al.⁶ in regard to reframing, and found that an increased use of emotional support and a decrease in self-blame positively affect PPC. Other psychological factors were investigated by Ho et al. who found that hope, optimism, and PPC are all positively correlated⁷. However, only hope was a significant individual indicator of PPC⁷.

The aim of this study was to further examine the relationships between biomedical variables, HRQoL, social factors, and subjective reports of PPC following treatment for HNC. It was hypothesized that a greater disease adversity overcome (survived), fewer disease and treatment side-effects, and higher HRQoL would be associated with greater PPC.

Methods

This was a prospective study using self-completion psychometric measures.

Participants

Ethical review was sought and granted. Potential participants were identified through a regional health informatics database. A questionnaire battery was sent via the mail, with a freepost return envelope, to all potential participants. No follow-up letters were sent.

To be approached as a potential participant, the person had to be over the age of 18 years and to have an understanding of English judged by clinical staff to be sufficient to complete a series of questionnaires in English. Their tumour had to have a histological diagnosis of squamous cell carcinoma (SCC) and be sited in the mouth, lip, oral cavity, salivary gland, pharynx, nasal cavity, or sinuses.

Potential participants were between 3 and 12 months post treatment and disease-free. The time frame of greater than 3

months post treatment was selected to allow for the acute effects of treatment to resolve and the demands of treatment (e.g. fatigue, travel, financial burden, family upheaval) to have lessened.

Of the 82 potential participants, 52 (63%; 36 male, 16 female) returned an at least partially completed questionnaire pack. Demographic data included age at time of diagnosis, sex, Index of Multiple Deprivation (IMD); UK government study of deprived areas in local councils based on income, employment, health, disability, education, skills and training, barriers to housing and services, crime, and the living environment¹⁴, and family status (married, living with partner, living alone, living with relative/friends). Medical data included tumour stage, date of diagnosis, treatment regimen, and date of treatment completion. Treatment regimen was split into three categories: surgery ($n = 16$), surgery and radiotherapy ($n = 17$), radiotherapy with or without chemotherapy (no surgery) ($n = 18$).

The Mann-Whitney *U*-test was used to compare medical (tumour stage, time since treatment, treatment regimen) and demographic (age at time of diagnosis, gender, family status, IMD) data between responders and non-responders to the questionnaires, and no significant difference was found between them. Table 1 provides demographic information of the respondents.

Questionnaires

The Silver Lining Questionnaire (SLQ) is a 38-item measure using a five-point Likert scale that examines the extent to which people believe their illness has resulted in a positive psychological change despite the negative consequences of being ill^{15,16}. The SLQ has not been used to investigate PPC in people specifically with or following HNC, other than in unpublished literature by the present authors. The SLQ has been used with mixed cancer cohorts (breast, colorectal, gynaecological, and lung)¹⁷. An additional strength of the SLQ is that it was developed in the same geographical region of the UK as where this research study was undertaken¹⁵.

The University of Washington Quality of Life Questionnaire (UW-QOL) version 4, specific for head and neck cancer, has 12 individual domains: pain, appearance, activity, recreation, swallowing, chewing, speech, shoulder function, taste, saliva, mood, and anxiety, and two sub-scales of physical function and social function¹⁸. The UW-QOL has been validated by comparison to the Karnofsky scale and Sickness Impact Profile, demonstrating an average criterion validity of 0.85¹⁸.

Table 1. Psychosocial characteristics of participants.

	Number	Mean	SD
Index of Multiple Deprivation	46	18.88	10.74
Gender			
Male	36		
Female	16		
Age at diagnosis, years	51	64.54	10.34
Family status			
Married/living with partner	35		
Living alone	8		
Living with relatives/friends	1		
Cancer stage			
I	10		
II	1		
III	13		
IV	26		
Treatment regimen			
Surgery	16		
Surgery + radiotherapy	17		
Radiotherapy + chemotherapy (no surgery)	18		
Months since treatment	52	6.52	2.80
SF-12 domains			
Mental component scale	26	41.68	10.00
Physical component scale	26	38.99	8.42
SLQ	52	11.85	9.46
UW-QOL – total	49	885.00	191.96
UW-QOL – physical function sub-scale	52	71.54	18.92
UW-QOL – social function sub-scale	52	75.40	16.78
UW-QOL domains			
Pain	52	75.96	23.35
Appearance	52	77.46	20.98
Activity	52	70.33	23.33
Recreation	52	74.31	23.71
Swallowing	52	76.98	23.74
Chewing	52	68.41	32.04
Speech	51	85.82	19.10
Shoulder	50	84.25	27.29
Taste	51	61.66	33.77
Saliva	50	61.84	35.72
Mood	51	75.00	23.49
Anxiety	51	72.39	24.78

SD, standard deviation; SF-12, Short Form 12; SLQ, Silver Lining Questionnaire; UW-QOL, University of Washington Quality of Life Questionnaire.

The medical outcomes Short-Form 12 (SF-12) is a generic health-related quality of life questionnaire with 12 items¹⁹. Results for each patient are expressed in terms of two meta-scores: the physical component summary (PCS) and the mental component summary (MCS). The SF-12 was selected over other longer versions or questionnaires in order to keep the total number of questions the respondents were asked to answer to a minimum.

Analysis

Baseline models of PPC were assessed. Linear mixed-effect models were used to assess effects of demographic, medical, and psychosocial variables on SLQ scores at baseline, and random coefficient models were used to assess effects of these variables on PPC scores. Separate models were run for total PPC score and each domain score. Time was calculated as

months since diagnosis and was included in the model using both linear and quadratic terms. The intercept and time slope were included as random effects in the models.

Predictor variables included in the model were socio-demographic (age, marital status) and medical (cancer stage) characteristics. Beta coefficients and standard errors were estimated using models containing all covariates listed earlier, and least squares mean estimates for SLQ total score were calculated from the models with covariates set to their mean levels. IBM SPSS Statistics version 19.0 (IBM Corp., Armonk, NY, USA) was used for all analyses.

Results

The mean age of the participants at the time of diagnosis was 64.69 years (standard deviation 10.28), with an average age at time of completing the questionnaires of

65.63 years (standard deviation 10.31). Table 1 summarizes the descriptive data for all bio-psychosocial variables of participants completing the measures at between 3 and 12 months. The table provides data on the sample size of each variable, including sub-categories of variables such as the four categories of cancer stage.

Table 2 shows the results of the linear mixed-effects model with SLQ as the dependent variable. This modelling was split into four sections to allow for the number of responses per variable to not exceed the rule of thumb of 10 responses per variable of Kleinbaum et al.²⁰. The first included modelling with IMD, gender, age at diagnosis, and family status. The sentence should therefore read: The second included modelling cancer stage, with stage II and III amalgamated, treatment regime and time since treatment (Table 2). The third section included modelling the SF-12 with the mental and physical component scales, but not the other sub-scales due to the small number of respondents. The fourth section included modelling with the total UW-QOL without the sub-scales for the same reason as not including the SF-12 sub-scales.

The results show that at between 3 and 12 months after the completion of treatment, family status, stage of the tumour, and the treatment regimen all had a relationship with PPC as defined with the SLQ ($P = 0.050$, $P = 0.006$, and $P = 0.044$, respectively). Figure 1 shows that between 3 and 12 months post-treatment, participants with low stage tumours (stage I) had a higher reported level of PPC than those with stage II and III tumours and noticeably higher PPC than those with stage IV tumours. In the same time frame, participants who had undergone surgery alone reported more positive changes than those who had undergone surgery with radiotherapy, and than those who were not treated surgically but who had radiotherapy with or without chemotherapy.

'Time since completion' was entered into the model as raw interval data (unlike data used in the longitudinal model where time was categorized into groups) and did not show any relationship with change.

Figure 2 shows how family status was associated with PPC (SLQ total). For the period covered by this analysis, it was found that being married or living with a partner rather than living alone was associated with a greater level of PPC (Table 2).

Discussion

The aim of this study was to investigate patient reports of PPC in relation to HRQoL at 3 to 12 months following the

Table 2. Association of demographic, medical, and psychosocial characteristics with SLQ scores.

Covariate	Baseline model	
	Estimate (SE)	P-value
Index of Multiple Deprivation	8.706 (16.941)	0.114
Gender	0.128 (6.108)	0.321
Age at diagnosis	8.637 (18.590)	0.257
Family status		0.050
Married/living with partner	-7.600 (9.428)	
Living alone	-18.125 (9.860)	
Living with relatives/friends	- ^a	
Cancer stage		0.006
I	11.900 (3.104)	
II and III combined	50.090 (9.437)	
IV	-0.804 (3.209)	
Treatment regimen		0.044
Surgery	6.989 (3.104)	
Surgery + radiotherapy	22.764 (3.056)	
Radiotherapy + chemotherapy (no surgery)	- ^a	
Time since treatment	14.549 (25.252)	0.806
SF-12 domains		
Mental component scale	91.333 (74.573)	0.634
Physical component scale	91.333 (74.573)	0.634
UW-QOL - total	57.000 (30.468)	0.231
UW-QOL - physical function sub-scale	10945.6 (2544.8)	0.198
UW-QOL - social function sub-scale	12710.7 (2709.9)	0.488

SE, standard error; SF-12, Short Form 12; SLQ, Silver Lining Questionnaire; UW-QOL, University of Washington Quality of Life Questionnaire.

^a Covariance parameter is redundant.

completion of treatment for HNC. A post-al survey of patients with HNC had a good response rate (63%), which is comparable to other studies^{21,22}.

Linear mixed-effects models suggested that both the clinical stage of the tumour and the treatment regimen undergone by the person had a relationship with PPC as defined with the SLQ. People with stage I tumours had a higher reported level of PPC than those with stage II and III tumours, and patients with stage I-III tumours had higher PPC than those with stage IV tumours. This may be because people diagnosed with a low stage (I or tumour in-situ) did not interpret this as a significant life-changing event. People diagnosed with stage IV tumours may have experienced such significant distress, or negative treatment side effects, that they were not able to find any positive change, at least up to 1 year post treatment. This may change in the long term, but has yet to be investigated in an HNC population.

Respondents who had surgery alone reported more PPC than those who had surgery with just radiotherapy and those who had radiotherapy with or without

Silver Lining Questionnaire against Cancer Stage between 3 and 12 months

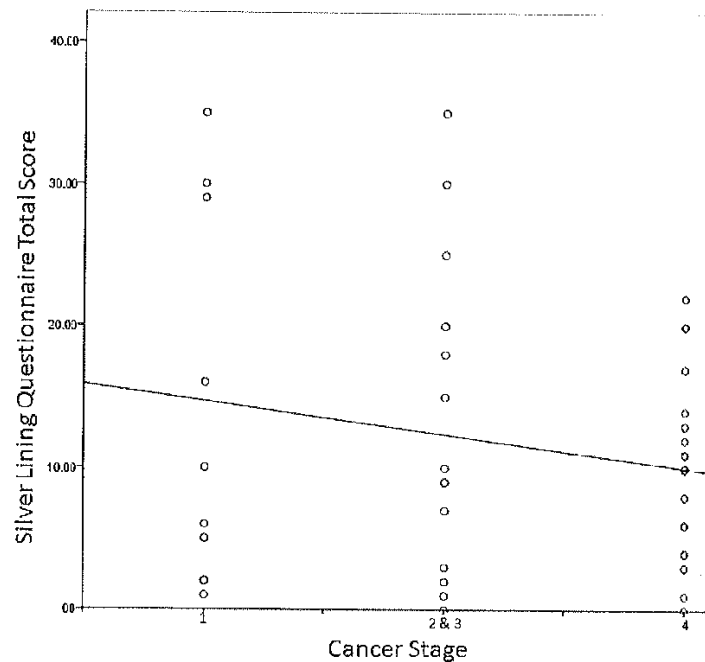


Fig. 1. Relationship between the Silver Lining Questionnaire (SLQ) score and cancer stage.

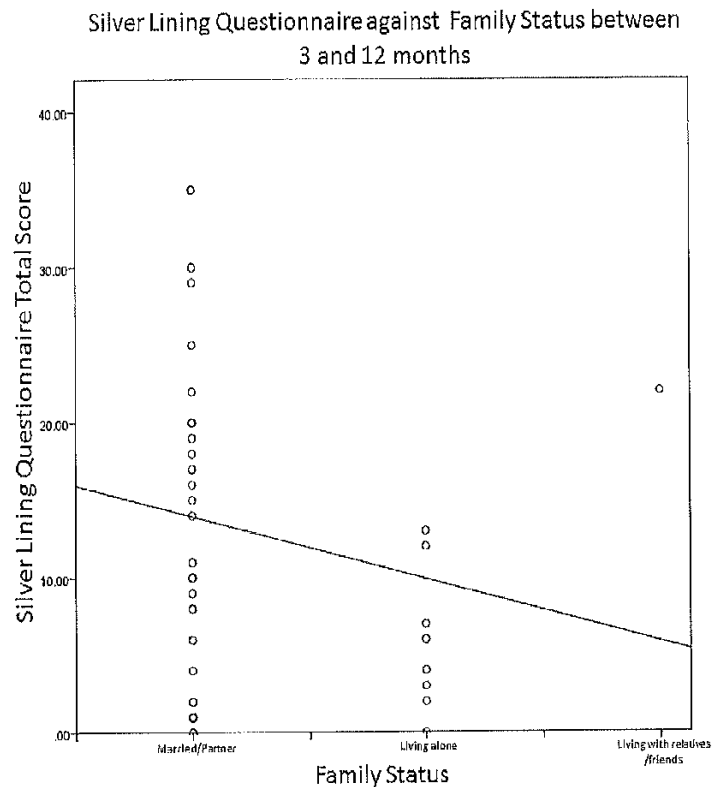


Fig. 2. Relationship between the Silver Lining Questionnaire (SLQ) score and family status.

chemotherapy (no surgery). There is an interrelationship between tumour staging and treatment regimen that may also impact PPC. Lower stage tumours, i.e. smaller, locally defined (no invasion into other tissues, or metastasis), will receive less aggressive curative treatments. People who are treated with surgery alone, while still receiving the diagnosis of cancer and undergoing the same diagnostic investigations as those people who have radio- and chemotherapy, are likely to have surgery as a one-off event with a minimal hospital stay time. Many surgical interventions do not require multiple hospital visits to receive treatment. On completion of the surgery, people with low stage tumours may receive a clear report from the surgeon that they could remove the entire tumour (if the surgery does not fully clear the tumour, these people usually go on to receive radiotherapy). This acute experi-

ence of a cancer diagnosis and treatment along with reassurance from the surgeon may mean that a person does not perceive the experience as traumatic enough to change their perceptions of self and how they relate to others.

Using the IMD to measure socio-economic status showed that, in the short term (3–12 months), there was no relationship with PPC. This differs from research undertaken in people with breast cancer, where those who were more deprived had more PPC²³. It is unclear why these differences may exist, but it may be that the IMD is not sensitive enough to show a change or that the sample was not large enough. It may also be that people with a higher socio-economic status are more likely to return the measures and may be less worried about financial matters in the short term and subsequently are able to develop PPC.

A social factor that was found to be related to higher levels of PPC in this short time frame (3–12 months) was being married when compared to living alone, and this in turn was more beneficial than living with a partner or relative(s). This supports the work of Ho et al., who found that oral cancer patients who were married reported higher levels of PPC than those who were unmarried⁷. Having close social relationships with family and friends is a key trait of PPC²⁴.

No psychological variables (collected using the SF-12 and UW-QOL) showed an association with PPC. This differs from the results reported in breast cancer studies. Danhauser et al. suggested that PPC is mediated in people with a 'high' quality of life and/or mental health²⁵. These HRQoL factors may be affected by the passage of time and overcoming or adaptation to the side effects of treatment. However, the

time since completion of therapy in the short term (between 3 and 12 months) did not show any relationship with PPC. These findings suggest that PPC within an HNC population might be affected by certain demographic, medical, and psychological factors in the short term (3 to 12 months).

In treatment and research on long-term cancer survivorship, a follow-up of at least 5 years after diagnosis is typical. The patients in the current study were assessed at between 3 and 12 months post treatment. Consideration should be given to the changes and experiences that people may encounter in the extended timeframe, including other stressful events and concurrent diseases. Additionally, there is evidence that positive changes may sometimes represent biased, self-enhancing, and self-protecting illusions rather than actual improvements^{26,27}. Some reports of growth are likely to reflect actual change that can be linked to behaviour, whereas other reports of growth may represent cognitive distortions that individuals make in their efforts to cope with distress²⁸.

No one would disagree that on first consideration a diagnosis of cancer is traumatic; however cancer is not a discrete, singular stressful experience. Rather it entails a cascade of potential stressors, from diagnosis, treatment, and treatment side effects to ongoing concerns of recurrence²⁸. This raises the question as to what is the trauma. Are there multiple traumas experienced by people diagnosed with cancer? Unlike an acute trauma, where the likelihood of re-experiencing the same event is low, Humphries et al. found that patients with HNC may think continuously about what might happen, with the fear of recurrence, "waiting for the other shoe to drop"²⁹. So what an individual identifies as the trauma is a challenge to the investigation of PPC.

Further investigation of PPC may benefit from the identification of a single traumatic time point in the cancer journey, or the participant identifying compounding events on their trauma journey, the summation of which represents a traumatic event. Clinical experience has shown that patients attending clinics may fear or suspect a diagnosis of cancer and the confirmation of a cancer diagnosis is in some part a relief. Furthermore, with the long-term side effects of treatment such as radionecrosis, xerostomia, or trismus, they may not feel that they have reached the end of their cancer journey. In these ways, cancer patients are never really 'post trauma', and further longitudinal investigations into the development of PPC in

general and specifically in people who have experienced HNC would be beneficial.

Funding

No funding was sought to undertake this research.

Competing interests

The authors have no competing interests.

Ethical approval

Ethical approval was obtained and issued from NRES Committee East Midlands — Northampton (reference number 167514 - 15/EM/0052).

Patient consent

Not required.

References

1. Cancer Research UK. *Oral cancer incidence statistics*. 2014. <http://info.cancerresearchuk.org/cancerstats/types/oral/incidence/> [Accessibility verified February 7, 2017].
2. Rogers SN, Fisher SE, Woolgar JA. A review of quality of life assessment in oral cancer. *Int J Oral Maxillofac Surg* 1999;28:99–117.
3. Joseph S, Linley PA, Andrews L, Harris G, Howle B, Woodward C, Shevlin M. Assessing positive and negative changes in the aftermath of adversity: psychometric evaluation of the changes in outlook questionnaire. *Psychol Assess* 2005;17:70–80. <http://dx.doi.org/10.1037/1040-3590.17.1.70>
4. Sears SR, Stanton AL, Danoff-Burg S. The yellow brick road and the emerald city: benefit finding, positive reappraisal coping, and posttraumatic growth in women with early stage breast cancer. *Health Psychol* 2003;22:487–97. <http://dx.doi.org/10.1037/0278-6133.22.5.487>
5. Calhoun LG, Tedeschi RG. Posttraumatic growth: future directions. In: Tedeschi RG, Park CL, Calhoun LG, editors. *Posttraumatic growth: positive changes in the aftermath of crisis*. Mahwah, NJ: Lawrence Erlbaum Associates; 1998. p. 215–38.
6. Harrington S, McGurk M, Llewellyn CD. Positive consequences of head and neck cancer: key correlates of finding benefit. *J Psychosoc Oncol* 2008;26:43–62.
7. Ho S, Rajandram RK, Chan N, Samman N, McGrath C, Zwanen RA. The roles of hope and optimism on posttraumatic growth in oral cavity cancer patients. *Oral Oncol* 2011;47:121–4. <http://dx.doi.org/10.1016/j.oraloncology.2010.11.015>
8. Holmaat K, van der Spek N, Cuijpers P, Leemans CR, Verdonck-de Leeuw IM. Post-traumatic growth among head and neck cancer survivors with psychological distress. *Psychooncology* 2016;26:96–101. <http://dx.doi.org/10.1002/pon.4106>
9. Llewellyn CD, Horney DJ, McGurk M, Weinman J, Herold J, Altman K, Smith EE. Assessing the psychological predictors of benefit finding in patients with head and neck cancer. *Psychooncology* 2013;22:97–105. <http://dx.doi.org/10.1002/pon.2065>
10. Leong AM, Nik Jaafar NR, Zakaria H, Rajandram RK, Mahadevan R, Mohamad Yunus MR, Shah SA. Posttraumatic growth, depression and anxiety in head and neck cancer patients: examining their patterns and correlations in a prospective study. *Psychooncology* 2015;24:894–900. <http://dx.doi.org/10.1002/pon.3740>
11. Harding S, Sanipour F, Moss T. Existence of benefit finding and posttraumatic growth in people treated for head and neck cancer: a systematic review. *PeerJ* 2014;2:e256. <http://dx.doi.org/10.7717/peerj.256>. eCollection 2014.
12. Low CA, Stanton AL, Thompson N, Kwan L, Ganz PA. Contextual life stress and coping strategies as predictors of adjustment to breast cancer survivorship. *Ann Behav Med* 2006;32:235–44. http://dx.doi.org/10.1207/s1532796abm3203_10
13. Manne S, Ostroff J, Winkel G. Posttraumatic growth after breast cancer: patient partner, and couple perspectives. *Psychosom Med* 2004;66:442–54.
14. Communities and Local Government The English indices of deprivation 2007. <http://webarchive.nationalarchives.gov.uk/20100411141238/http://www.communities.gov.uk/communities/neighbourhoodrenewal/deprivation/deprivation07/> [Accessed August 12, 2017].
15. Sodergren SC, Hyland MB. What are the positive consequences of illness? *Psychol Health* 2000;15:85–97. <http://dx.doi.org/10.1080/08870440008409290>
16. Positive Psychology Center, University of Pennsylvania. Silver Lining Questionnaire download. <https://ppc.sas.upenn.edu/resources/questionnaires-researchers/silver-lining-questionnaire> [Accessibility verified June 18, 2017].
17. McBride O, Schroevens MJ, Ranchor AV. The structure of adversarial growth in a sample of cancer patients 8 years post-diagnosis: a revision of the SLQ-38. *Psychol Health* 2009;24:1197–213. <http://dx.doi.org/10.1080/08870440802108900>
18. Rogers SN, Gwanne S, Lowe D, Humphris G, Yueh B, Weymuller Jr EA. The addition of mood and anxiety domains to the University of Washington Quality of Life scale. *Head Neck* 2002;24:521–9. <http://dx.doi.org/10.1002/hed.10106>
19. Ware Jr J, Kosinski M, Keller SD. A 12-item short-form health survey: construction of

- scales and preliminary tests of reliability and validity. *Med Care* 1996;34:220-33.
20. Kleinbaum DG, Kupper LL, Muller KE. *Applied regression analysis and other multivariable methods*. Baltimore, MD: Wadsworth Publishing Co. Inc.; 1997.
 21. Millsopp L, Brandom L, Humphris G, Lowe D, Stat C, Rogers S. Facial appearance after operations for oral and oropharyngeal cancer: a comparison of case notes and patient-completed questionnaire. *Br J Oral Maxillofac Surg* 2006;44:358-63. <http://dx.doi.org/10.1016/j.bjoms.2005.07.017>.
 22. Vickery JF, Litchford G, Hewison J, Bellew M, Feber T. The impact of head and neck cancer and facial disfigurement on the quality of life of patients and their partners. *Head Neck* 2003;25:289-96. <http://dx.doi.org/10.1002/hed.10206>.
 23. Tonich PL, Helgeson VS. Is finding something good in the bad always good? Benefit finding among women with breast cancer. *Health Psychol* 2004;23:16-23. <http://dx.doi.org/10.1027/0278-6133.23.1.16>.
 24. Tedeschi RG, Calhoun LG. Posttraumatic growth: conceptual foundations and empirical evidence. *Psychological Inquiry* 2004;15:1-18. http://dx.doi.org/10.1207/s15327965pli1501_01.
 25. Danhauser SC, Russell G, Case LD, Sohl SJ, Tedeschi RG, Addington EL, Triplett K, Van Zee KJ, Naftalis EZ, Levine B, Avis NE. Trajectories of posttraumatic growth and associated characteristics in women with breast cancer. *Ann Behav Med* 2015;49:650-9. <http://dx.doi.org/10.1007/s12160-015-9696-1>.
 26. Park CL, Helgeson VS. Introduction to the special section: growth following highly stressful life events—current status and future directions. *J Consult Clin Psychol* 2016;74:791-6. <http://dx.doi.org/10.1037/0022-006X.74.5.791>.
 27. Taylor SE, Armor DA. Positive illusions and coping with adversity. *J Pers* 1996;64:873-98.
 28. Gurevich M, Devins GM, Rodin GM. Stress response syndromes and cancer: conceptual and assessment issues. *Psychosomatics* 2002;43:259-81. <http://dx.doi.org/10.1176/appi.psy.43.4.259>.
 29. Humphries GM, Rogers S, McNally D, Lee-Jones C, Brown J, Vaughan D. Fear of recurrence and possible cases of anxiety and depression in orofacial cancer patients. *Int J Oral Maxillofac Surg* 2003;32:486-91.

Address:
Sam Harding
Department of Health and Social Sciences
University of the West of England
Bristol
BS16 1QY
UK
Tel.: +44 (0)7944 363973
E-mail: sharding.jb@gmail.com

3.2.4 Study limitations

3.2.4.1 Study design

The most often cited limitation of single time point studies using a cross-sectional design is that they cannot answer questions about the stability of a characteristic or process over time (Miller, 2007). A further criticism of cross-sectional studies is that their external validity (i.e. generalisability) may be affected by historical/cultural differences between cohorts (Achenbach, 1978). This is perhaps their most serious limitation; you cannot easily separate the effects of age from the effects of belonging to a particular cohort, especially if that cohort is defined by birth. Miller (2007) argues that the seriousness of this problem relates to the dependent variable: the more basic or biological the variable (e.g. heart rate, visual acuity), the less likely it is that the cohort effect will be present. Adopting a cross-sectional approach to studying the process of change or development would not permit the identification of predictors associated with having a reduced ability to speak or eat, whereas adopting a longitudinal approach would allow such analyses (Campbell, 1988).

3.2.4.2 Participant response rates

The scales used in the paper 2 had completion rates between 48 and 95 percent. Information about progressive health issues not related to cancer was not presented in the electronic records interrogated in this research. Therefore, the responses sent back providing this information were very useful, and highlight that future longitudinal research needs to record comorbidities and significant life events that

may interact with primary health condition in the development or moderation of PPC.

The three measures used (Medical Outcomes Short Form -12 (SF-12), Silver Lining Questionnaire (SLQ), University of Washington (UoW)) were ordered using a Latin Square. Appendix 4 provides great detail about each of these measures. The ordering of the presentation of the questionnaires means it is unlikely that the poor completion rate is due to fatigue related to the length of the survey. In the research connected with paper 2 there was a surprisingly poor completion rate for the SF-12 (48%).

It is possible that the SF-12 may have a low level of acceptability for respondents. There has previously been concern over completion rates for the SF-12 in community studies. An Australian study found that 78% completed all 12 items (Lim and Fisher, 1999), and a British study found 84.1% of 55,000 respondents completed all 12 items (Jenkinson *et al.*, 2001), but the research undertaken in the production of paper 2 is notably worse than these. No free-text was asked for from the respondents regarding incomplete returns, however, the reason for the high incompleteness in the larger project remains unclear and would benefit from discussion with my public patient involvement (PPI) group to try to identify possible reasons. The SF-12 remains effective as a brief but broad-ranging instrument, suitable for survey use and sensitive to change as an evaluative instrument (McDowell, 2006). Further investigation is required to understand the low completion rates in the wider project from which this paper is derived, but I would retain this measure in future studies/work due to its generic nature and brevity.

There is no data reporting on completion or partial completion rates where the respondents returned the SLQ via a postal survey. One study that recruited 194 patients had a 1.5% non-completion rate but did not report how many of their participants partially completed measures (Sodergren *et al.*, 2004). The SLQ authors describe a process to calculate a total score from a return that has at least 93% of the items completed. Previous studies have not reported the use of this process for calculating a total score from completed returned surveys. In research used in paper 2 there was a 71% total completion of the SLQ, with this rising to 95% using the correction method. Those who did not complete the scale at all reported that they “*did not understand*”, but it was not clear if it was the questions or the reason behind

being asked them. Others commented that they had not had an illness, or that the disease/treatment they had did not have any effect on their outlook on life.

The UoW is a disease-specific measure (HNC) and as such focuses on issues relevant to people who have had a diagnosis of this type of cancer. By virtue of its brevity, it does not include other issues related to QoL, e.g. cognitive function. The questionnaire has proven itself a suitable head and neck measure for routine clinical practice as it is quick and simple for patients to complete and is easy for clinicians and researchers alike to interpret (Rogers *et al.*, 1998). A completion rate of 88% was reported in a study where the measure was compared to QoL measures (Rogers *et al.*, 1998). In this work leading to paper 2, 93% of the returns had a fully completed UoW. The high level of completion indicates a good level of acceptability of the questions and recognition that the measure is directly related to the person's experiences of HNC and its potential acute and long term consequences.

The questionnaires used in the presented research were not the only ones that could have been used. But they were selected for use in the research that led to paper 2 for their psychometric properties, previous use in the patient group, and length. Even so, it might be that these measures do not ask the type of questions in a way that would elicit relevant information from the particular patient cohort. An elderly population such as the HNC cohort, who often have multiple comorbidities, may have found that even 25 minutes to complete all the measures too much of a burden.

3.2.4.3 Limitations

The response rate or non-response error refers to the condition wherein people of a particular ilk are systematically not represented because such people are alike in their tendency not to respond. There could be multiple groups of people who do not respond to surveys generically because such groups, by their very nature, are disinclined to respond (e.g., introverts, extremely busy people, people with low esteem; Krosnick, Lavrakas and Kim, 2013; Porter and Whitcomb, 2005). In the research leading to paper 2 there was no difference between responders and non-responders on the variables that we had for non-responders. There may be factors where they would differ, which may make it difficult to say how the entire sample would have responded. Generalising from the sample to the intended population thus becomes risky. For this reason, non-response error in mail surveys has long concerned social science researchers (Sivo *et al.*, 2006). However, real world issues

must be considered and it is not possible to measure every variable that might impact on a person's likelihood of responding. Therefore working with the information collected, I am confident that the respondents are representative of the people diagnosed with HNC in the local geographical area.

3.2.5 Contribution to knowledge and autoethnographic issues

3.2.5.1 Contribution to knowledge

Paper 2 found a relationship between bio-medical variables, Health Related Quality of Life (HRQoL), social factors, and subjective reports of PPC following treatment for HNC. We found that a greater disease adversity overcome (survived), fewer disease and treatment side-effects, and higher HRQoL were associated with greater PPC, as defined by the SLQ.

3.2.5.2 Autoethnographic issues

As with paper 1, Appendix 5 has an overview of how the research undertaken in paper 2 was disseminated prior to the paper's publication. It also provides a list of papers where paper 2 has been cited, and this is followed by a quality appraisal of the paper.

Early on in the research I established a public patient involvement (PPI) group. This group had 12 people who had all experienced HNC in the last 5 years. All had been part of previous research projects I had undertaken, so had a pre-existing relationship with me. We discussed the theoretical basis of the study and the potential methodologies that we could use. They helped construct the protocol, ethics forms and applications, as well as reviewing and commenting on materials before I disseminated them. Public Patient Involvement group's involvement should mean that the research and its findings were more relevant to the patients' experience. Their thoughts and feelings about their treatment journey and how research can fit into this and how it might make a difference in the treatment of future patients was fascinating and informed not only this project, but all my subsequent research and its design.

3.2.6. Presenting findings to an HNC population

A couple of years into the project that led to paper 2, I presented some initial findings at the 8th International Conference Quality of Life in Head & Neck Cancer. The title of the presentation was 'The impact of treatment for head and neck cancer

on posttraumatic growth'. The audience at this conference were HNC clinicians, Nurses, Physiotherapist and Speech and Language Therapists. There were however a number of people who had been treated for HNC. It was this cohort of people that provided me with an interesting learning experience and lots to think about in how I framed my future dissemination.

At the time of the presentation I was using the term 'Posttraumatic Growth', rather than PPC. I was more than half way through the talk when several people entered. At the end of the presentation during questions one of these people stood up and asked '*How dare I say that having HNC is a good thing!*'. I was taken aback! I attempted to answer the question, but not to the person's satisfaction. At this point the chair called the session to the end and I asked if I could speak to the gentleman if he had time.

It took some time, allowing the gentleman to have his say about how awful his diagnosis and treatment had been. He also stated that if I had ever spoken to anyone who had experienced HNC then there would be no way I would come up with such an 'idiotic' research idea.

It was a real challenge to start my side of the conversation with him. He felt that my acknowledgement of his trauma was 'lip service' to stop him complaining. Where I had to start was with an apology for his feelings, and with the hope that he would allow me to give him an overview of the start of my presentation that he had unfortunately missed; my explanation that for Posttraumatic Growth to occur there had to have been a trauma.

As well as the conceptualisation of a post traumatic psychological change, it transpired that a linguistic hurdle needed to be overcome. When we had unpacked his 'misunderstanding' and defused the situation, it transpired that he interpreted the word 'growth' in posttraumatic growth to be directly related to the growth or re-growth of the cancerous tumour. So how could the growth of the cancer ever be a good thing! This was a revelation, and I was very grateful for him sharing this interpretation with me. I explained how my PPI group had never mentioned this and that this might be because of the extended periods of time I had spent with them developing the work. We discussed changing terminology and acceptability of Posttraumatic Growth, and we thought that PPC would be a better phrase when talking to people who had experienced cancer as their trauma.

3.2.7 Future research questions

In summarising the findings of the study, an interesting pattern was found. Time since completion of therapy in the short term, between 3 and 12 months, did not show any relationship with PPC. However, some medical factors did show some association. The analysis suggested that stage of the tumour and the treatment regimen undergone by the participant both have a relationship with PPC as defined with the SLQ. Participants with stage 1 tumours reported a higher level of PPC than those with stage 2 and 3 and noticeably higher than those with stage 4 tumours. Participants who had surgery alone reported more PPC than those who had surgery with radiotherapy, and those who were not treated surgically but who had radiotherapy with or without chemotherapy. A social factor related with higher levels of PPC in this time frame was being married or living with a partner when compared to living alone or with relatives.

The question left was; is this pattern maintained over time? Do people in the short term report psychological benefit from surviving, but is this benefit actually an underlying reformation of the person's assumptive world, and thus a PPC?

3.3 Summary

3.3.1 Summary of contribution to knowledge

Paper 1, presents the first study to assess the presence and prevalence of PTSD in a COPD population. Whilst paper 2, built on a very limited number of publications investigating PPC following HNC. It found a relationship between biomedical, HRQoL and social factors in the development of PPC within a year of completion of treatment.

3.3.2 Summary of autoethnographic issues

Both underlying pieces of research involved working with people with the health condition of interest, to design, refine, deliver and disseminate the work. I enjoy working with PPI groups, as it ensures the relevance of the research to the people affected by the disease. Building long term relationships where PPI group members feel able to freely voice their experiences and their thoughts about the research projects, was invaluable, and had a huge impact on my ongoing development and future research practices.

Chapter 4 Pre & Post Studies

This chapter examines the conduct of primary research investigating the impact on Quality of Life (QoL) of a health intervention. This is a more complex research question than the identification of an impact as described in the previous chapter, as it looks at the impact of an intervention on a person, rather than if a phenomenon exists within them. The use of pre- and post- measures can support correlations and suggest where a causal link between the health intervention and change in QoL may exist.

This pre- post- approach is reported in three papers focusing on patient groups who experience either pulmonary rehabilitation (PR, paper 3) or Hyperbaric Oxygen Therapy (HBOT, papers 4 and 5). These groups completed both generic and disease specific QoL measures before and after the intervention. This allowed us to identify the clinical and psychological impact of an intervention on a patient cohort.

4.1 Paper 3 - Clinical experience leading to the research work and production of the paper

While working in the Respiratory Research Unit, primarily to undertake the work reported in paper 1 (Chapter 3), I had the capacity to support other research projects. One was the development and validation of the Lung Information Needs Questionnaire (LINQ). This gave me the opportunity to undertake focus groups with people with a condition that I was not familiar with. This enabled me to ask patients to teach me, to allow them to be the experts in their own condition. My lack of experience meant that I was free of clinical bias and able to ask clarifying questions that may have been missed by clinicians.

The development and use of LINQ also fitted into my developing identity as a health psychologist. An element of this work was developing an understanding of the patients' understanding of Chronic Obstructive Pulmonary Disease (COPD), and how this has two elements; their knowledge and their information needs. At this time there were several COPD patient knowledge assessments, based either on questionnaires (open or closed questions) or using scenarios. However, patient information needs was an under-investigated area. The challenge with this work was to identify the needs that are relevant to the patients and clinicians, and that clinicians can use to improve the patient's treatments. This work situated itself

within the social cognitive models of health (e.g. Theory of planned behaviour; Ajzen, 1991) and further refined by health behaviour models and patients' understandings and beliefs about their illness (Leventhal, Nerenz & Steele, 1984; Leventhal, Diefenbach, & Leventhal, 1992).

4.1.1 Research Questions

This paper sought to:

1. Assess the ability of the LINQ to measure changing information needs before and after PR
2. Assess the variation in the LINQ score between sites
3. Investigate the relationship between the LINQ scores and other outcomes including QoL and exercise capacity

4.1.2 Study Design

Patient knowledge questionnaires are useful for assessing the effectiveness of educational programmes. This type of questionnaire varies in both content and length, reflecting variability in what clinicians believe COPD patients should know. The longer ones provide a more comprehensive form of assessment. For example, the Bristol COPD Knowledge Questionnaire has 65 items and takes about 20 minutes to complete (White et al, 2006). However, this comprehensiveness is achieved at the price of convenience for everyday clinical use. Patients differ in terms of depth and type of information that they seek, and so, whatever their length or content, such questionnaires can fail to reflect the patient perspective in terms of what the patient wants to know. What an individual patient wants to know is a reflection of the individual differences between patients and the fact that psychological problems, such as anxiety, depression and social isolation, affect the way patients seek and respond to education. Guidelines recommend that education should take into account the differing needs of patients at differing stages of their disease (National Collaborating Centre for Chronic Conditions, 2004). It is also important to understand that needs can differ from the perspective of the patient and the clinicians.

Information needs can be defined in two ways. First, if a patient expresses a desire for more information, then the patient has an information need, thereby taking into account differing levels of educational need. Second, a clinician can believe a

patient's response to a question suggests their self-management is compromised. This may also indicate that the patient has an information need. In contrast to knowledge questionnaires, clinician defined information needs provide a more focused perspective, identifying where lack of knowledge can compromise the patient's ability to self-manage. Such evidence includes research that smoking cessation and exercise affect prognosis and QoL, and that early response to symptoms reduces the impact of an exacerbation. An information needs questionnaire does not necessarily inform the clinician what the patient knows, but it does show that there is an aspect of education that needs attention, and because it is designed with brevity in mind is particularly suited for pre-intervention assessment.

Following the development of the LINQ and assessment of its test-retest reliability, we wanted to assess if it was able to measure changing information needs. To do this, the work presented in paper 3 sampled several groups of patients with COPD, who were participating in PR, to assess pre and post information needs. These changing information needs were explored in response to other factors such as QoL and exercise.

A prospective pre-, post-intervention cohort study design was adopted. A cohort study design was chosen because the primary research question, that of assessing the ability of the LINQ to evaluate information needs before and after PR, described a coherent group of individuals all experiencing the same phenomenon. While the study was spread across six sites, careful research design ensured that, for the purposes of analysis, they could be considered one cohort. Since patients were referred for PR, it was possible to gather pre-intervention data as well as post-intervention data.

4.1.3 Educational impact of pulmonary rehabilitation: Lung Information Needs Questionnaire



available at www.sciencedirect.com



journal homepage: www.elsevier.com/locate/rmed



Educational impact of pulmonary rehabilitation: Lung Information Needs Questionnaire

Rupert C.M. Jones^{a,*}, Xu Wang^a, Sam Harding^a, Julia Bott^b,
Michael Hyland^c

^a Respiratory Research Unit, Peninsula Medical School, Devon, 1 Davy Road, Plymouth PL6 8BX, United Kingdom

^b Respiratory Care Team, Surrey Primary Care Trust, Chertsey, TW15 3AA, United Kingdom

^c School of Psychology, University of Plymouth, Devon, PL4 8AA, United Kingdom

Received 18 December 2007; accepted 17 April 2008

KEYWORDS

Education;
COPD;
Information needs;
Pulmonary
rehabilitation

Summary

Introduction: The Lung Information Needs Questionnaire (LINQ) assesses, from the patient's perspective, their need for education. This questionnaire yields a total score and scores in six domains: disease knowledge, medicine, self-management, smoking, exercise and diet. The aim of this study was to assess the sensitivity of the LINQ to change before and after pulmonary rehabilitation (PR).

Method: PR programmes across the UK recruited 158 patients (male = 94; 59%). The participants completed the LINQ and other measures as used by the individual sites pre- and post-PR, including the Shuttle Walking Test, Chronic Respiratory Disease Questionnaire, the Hospital Anxiety and Depression Scale.

Results: Data were analysed on 115 patients who completed data collection pre- and post-PR. The LINQ total scores, and subscales scores across all sites improved significantly with large effect sizes, except for the smoking domain as information needs about smoking were well met prior to PR. There were similar patterns of information needs at baseline and after PR in all sites.

Discussion: This study shows that the LINQ is a practical tool for detecting areas where patients need education and is sensitive to change after PR. The quality of the education component of PR can be assessed using the LINQ, which could be considered as a routinely collected outcome measure in PR. The LINQ may also be a useful tool for general practitioners to assess their patients' educational needs.

© 2008 Elsevier Ltd. All rights reserved.

Introduction

Chronic obstructive pulmonary disease (COPD) is a progressive disabling respiratory disease. As the disease

* Corresponding author. Tel./fax: +44 1752 764 259.
E-mail address: rupert.jones@pms.ac.uk (R.C.M. Jones).

advances patients suffer with increasing symptoms, impaired health status, disabilities and a spiralling decline in mental and physical wellbeing.¹ The decline in lung function can be reduced by stopping smoking. However, decline in function and activity can be reduced or improved by adopting appropriate strategies such as increasing physical activity,¹ pulmonary rehabilitation (PR) and self-management of exacerbations.^{2,3} Health status may be improved by drug treatment and PR.¹

As with any chronic life-threatening illness, patients need to understand the cause and nature of the problem, how treatment can help and what they can do for themselves to minimise its impact.⁴ In particular COPD patients' behaviour is important in determining their prognosis (e.g. smoking cessation) and disability (exercise). The starting point of changing behaviour is to impart information⁵; one of the most effective way to deliver this is through PR. PR is a programme of exercise and education delivered to groups of patients with chronic lung disease. It is of proven effectiveness in improving quality of life, exercise tolerance and reduction in dependency of patients with symptomatic COPD.⁵ The NICE guidelines recommend that PR should be offered to all patients who consider themselves functionally impaired by COPD.⁶ Currently assessment of changing health status and exercise capacity after PR is recommended, but not assessment of the education component.⁵ Education may be measured using knowledge or information needs questionnaires. COPD knowledge questionnaires^{8,9} tend to be long and predicated on the clinician's perspective as to what patients should know. Information needs are predicated from the patients' perspective, but until recently there have been no validated tools to measure them.

The Lung Information Needs Questionnaire (LINQ) (www.linq.org.uk) is a new tool which assesses, from the patient's perspective, the information they need to adequately understand their lung disease and to maximise their self-management skills. The LINQ is a self-complete questionnaire with 16 items and 6 subscales, and was designed using an iterative process involving 10 focus groups of invited COPD patients.¹⁰ The LINQ has been validated in cross-sectional data and for test-retest reliability, but not in terms of its sensitivity to change.¹⁰

Objectives

The aim of this study was to assess the ability of the LINQ to measure changing information needs before and after PR. Secondary aims were to assess (i) the variation in the LINQ scores between sites and (ii) the relationship between the LINQ scores and other outcomes such as quality of life and exercise capacity.

Materials and method

Ethical approval was obtained from the Central Office for Research Ethics Committees (COREC). Approval for each site was obtained from the relevant Local Research Ethics Committee and Primary Care or Hospital Trust.

Subjects

Patients with COPD who had been referred to PR were invited by letter to take part in the study. Written informed consent was obtained at the start of the PR programme. The study inclusion criteria were: physician diagnosis of COPD with a forced expiratory volume in one second (FEV₁) less than 80% of predicted and ratio of FEV₁ to forced expiratory vital capacity (FVC) less than 70%;⁶ able to attend regular community or hospital based PR, and provision of written informed consent. Patients were excluded if they had a serious literacy problems or a poor understanding of the English language.

Setting

Data were collected from patients attending PR at six sites across the UK — four from primary care and two from secondary care. Information relating to each programme was obtained from the study sites including setting, start and finish dates, frequency and duration of sessions, disciplines of staff involved, topics covered in the educational sessions, and details of the PR assessment tools. The education component in the different programmes involved key themes including the causes of COPD, the patho-physiology of COPD and treatment; including drugs, relaxation, exercise, nutrition and self-management. The disciplines involved in delivering the education are shown in Table 2.

Participants were asked to complete LINQ in addition to the usual assessments performed at the beginning and end of their PR programme. In the case of participants who completed the pre-study LINQ, but did not complete the PR programme, data were used for a summary of information needs only.

Measuring instruments

The main outcome measure for this study was the LINQ. The LINQ is a self-complete, tick box questionnaire with 16 questions that take an average of 6 min to complete. Each question has a multiple choice format and these are¹⁰ scored so that 0 = no information with increasing numbers depending on the level of need (the number of response choices varies between questions). The scores are summed for each domain and for the total score. There are six domains each with its own range of scores; disease knowledge (0–4), medication (0–5), self-management (0–6), smoking (0–3), exercise (0–5) and diet (0–2). The higher the score the greater the information requirements of the respondent. The total or global score (sum of all items, scores vary between 0 and 25) of the LINQ provides an overview of the patient's information needs and the individual domain scores identify their specific information needs. Full scoring details are available at <http://www.linq.org.uk>. To date a minimum significant clinical difference has yet to be established, but difference is less important than the final score achieved. Ideally, patients should have no information needs, but a score of 1 on any domain might be considered acceptable.

All six PR sites used the Hospital Anxiety and Depression Scale¹¹ (HADS) and Shuttle Walking Test¹² (SWT). Four sites used the Self-Reported version of Chronic Respiratory Questionnaire (CRQ-SR) which has four domains: dyspnoea, fatigue, emotional function and mastery.¹³ One site used the Clinical COPD Questionnaire¹⁴ (CCQ) which has three domains: symptoms, function, and mental state, and two sites used the St. George's Respiratory Questionnaire¹⁵ (SGRQ). All the questionnaires and assessments were conducted initially at pre-PR assessments and then during the final PR session.

Analyses

Data were analysed using the Statistical Package for the Social Sciences (SPSS, V14). Descriptive statistics were used to describe patients' characteristics. Changes in mean values on all total and subscale scores before and after rehabilitation programme were compared with paired *t*-tests (two-tailed).

In the absence of recognised clinically important changes for all the study variables, effect sizes were calculated for statistically significant changes using the following formula: effect size $r = \sqrt{(t^2/t^2 + df)}$. Boundaries recommended by Cohen (1988)¹⁶ were used to determine small (0.10), moderate (0.30), and large (0.50) changes in study variables. As a further examination of response to individual questions of the LINQ, McNemar's test was used to examine the changes of dichotomous responses to question

8 (i.e., whether patients had been provided with written instructions on how to deal with worsened breathing) and question 9 (i.e., whether patients had been told when to call an ambulance when breathing worsened).

The Pearson's correlation was used to examine the relationship between the LINQ scores and other outcome measures.

"Repeated measures analysis of variance was conducted using time (before and after PR) as a within-subject factor and different sites as a between-subject factor. This was performed to test the effect of PR across different sites."

Results

A total of 158 subjects (male = 94; 59%) were recruited. One hundred and twenty-six patients completed their PR programme. Eleven of the 126 had missing data and were removed from analysis. Thus, the analysis was based on 115 patients' data (male = 73; 63%). The average age was 69 years (SD = 8.55, range = 45–87 years).

The vast majority of subjects were smokers or ex-smokers. The overall mean exposure was more than 40 pack years. Disease severity characteristics for patients who completed the PR programme and those who did not are shown in Table 1.

Table 2 presents information on the PR programme and the number of patients recruited by each site. Baseline total and domain LINQ scores were similar in all sites.

Table 1 Patient characteristics: patients who completed the PR programme and with complete data and patients who dropped out of the PR programme

		Patients complete PR and with complete data (N = 115)		Patients who dropped out PR (N = 32)	
		Mean (SD)		Mean (SD)	
Age		69 (8.6)		70 (9.2)	
Pack years		41.1 (26.5) (range 0–175)		43.0 (26.7) (range 0–95)	
Smoking status ^a		N	%	N	%
Never smoked		9	8	1	3
Ex smoker		87	76	21	73
Current smoker		19	16	7	24
FEV1% predicted ^b	GOLD COPD category	N	%	N	%
50%–80%	II: Mild	39	41	4	21
30%–50%	III: Moderate	37	39	11	58
<30%	IV: Severe	18	20	4	21
MRC Dyspnoea Scale Baseline Score ^c		N	%	N	%
1		11	12	1	5
2		17	19	3	15
3		29	32	6	30
4		26	28	9	45
5		8	9	1	5

^a N = 115 for patients who completed PR and N = 29 for patients who dropped out.

^b N = 94 for patients who completed PR and N = 19 for patients who dropped out.

^c N = 91 for patients who completed PR and N = 20 for patients who dropped out.

Table 2 Summary information of the PR programme in each site

Site	Number of sessions per week	Duration of each session	Other assessment tools	Staff involved	Participants from each site who had completed LINQ data (N)
South Devon	1	2	HADS, CRQ-SR/CCQ, SWT and SBPQ	RPT, RNS, GP, fitness instructor, counsellor	23
Wiltshire	1	2	HADS, SGRQ, SWT	RPT, RNS, GP, psychologist, expert patient	20
Cardiff	3	2.25	SGRQ, HADS, SWT	RPT, physician, OT, dietician	26
London	1	2	CRQ-SR, HADS, SWT	RPT, RNS, OT, psychologist, Fitness instructor, Dietician, Doctors	12
Surrey	2	2.5	CRQ-SR, HADS, SBPQ, SWT	RPT, OT, RNS, dietician	28
Honiton	2	2.5	CRQ-SR, HADS, SWT	RPT, practice nurse, OT, psychologist	6

Note: CCQ = Clinical COPD Questionnaire; CRQ-SR = Self-Reported Version of Chronic Respiratory Questionnaire; HADS = Hospital Anxiety and Depression Scale; SBPQ = Short Breathing Problem Questionnaire; SGRQ = St. George's Respiratory Questionnaire; SWT = Shuttle Walking Test; RPT = Respiratory Physiotherapist; OT = Occupational Therapist; RNS = Respiratory Nurse Specialist

Changes in LINQ post-PR

In all sites, the LINQ total score post-PR improved significantly with a large effect size [$t(114) = 11.83$, $p < 0.001$, $r = 0.74$] (Table 3) and was similar across sites [$F(5, 109) = 2.03$, ns]. All domain scores improved significantly, with a medium to large effect size, with the exception of smoking in which baseline information needs were already well met. The diet and exercise domains improved the most, followed by self-management, disease knowledge and medicine domains (see Fig. 1). A repeated measures analysis of variance with time showed no significant interaction of time and sites.

Changes in the self-management domain

From Table 3 and Fig. 1, it can be seen that the baseline score for self-management and diet was the highest among the six domains of LINQ. Even after PR, information needs for self-management had not been well met.⁶ Before PR, 84 out of 115 patients (73%) reported that they had not been told when to call an ambulance if their breathing became worse. After PR, this figure dropped to 41/115 (36%). This change was statistically significant (McNemar's test, $p < .001$) indicating that a significantly greater proportion

of patients had been told when to call an ambulance with worsened breathing after PR. Similarly, after PR, a significantly greater proportion of participants had been provided with written instructions on how to deal with worsened breathing (McNemar's test, $p < .05$).

Changes in other outcome measures

After PR, there were significant improvements in SWT, HADS anxiety, HADS depression, CRQ-SR total and all four subscale scores. Improvements were also seen in CCQ total, SGRQ scores and SBQ scores but few of these reached statistical significance and this may be related to small sample size (Table 4).

The relationship between the LINQ scores and other outcome measures

The Pearson's correlation (r) did not reach significance between the total and domain scores of the LINQ and the pre-PR scores on secondary outcomes measures including: dyspnoea (MRC Dyspnoea Scale), SWT, HADS Anxiety, and HADS depression scores. Furthermore no significant correlations were found between changes in LINQ total and

Table 3 Mean scores, standard deviations and effect sizes of LINQ total and subscales over time ($N = 115$)

	Pre PR	Post PR	Significance	Effect size
	Mean (SD)	Mean (SD)	t	r
LINQ total score	9.6 (3.6)	5.1 (2.9)	11.83**	0.74
Disease Knowledge	1.7 (1.0)	1.1 (0.8)	6.49**	0.52
Medicines	0.7 (0.9)	0.3 (0.6)	5.20**	0.44
Self-management	3.6 (1.6)	2.2 (1.3)	7.74**	0.59
Smoking	0.2 (0.5)	0.2 (0.4)	1.07	ns
Exercise	2.1 (1.4)	0.8 (1.1)	8.85**	0.64
Diet	1.4 (0.7)	0.6 (0.7)	9.66**	0.67

Note: ** $p < .001$. Effect sizes (r): small (0.10), moderate (0.30), and large (0.50)

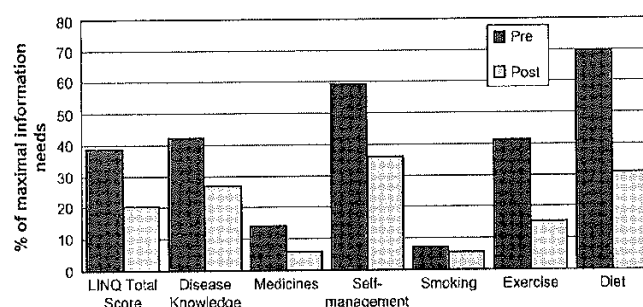


Figure 1 The mean LINQ domain and total scores pre and post PR expressed as a percentage of maximum possible information needs. Note: The greater the score the higher the information needs.

domain scores and changes in dyspnoea, SWT and HADS domain scores, except for that between change in medicine domain scores and change in SWT scores ($p = 0.04$). If a Bonferroni correction were applied, there would be no significant correlations.

Discussion

In this study, we investigated the changes in LINQ following PR. We found that the LINQ to be an effective tool in detecting information needs both at baseline and post PR. The changes associated with PR were statistically significant and occurred with a large effect size in all sites.

All domain scores improved with the exception of smoking, where patients' information needs were already well met before PR. It is of note that information needs in the self-management domain were poorly met before and even after PR in that many patients were not given written action plans as recommended by guidelines.⁵ This indicates that PR programmes need to provide patients with written instructions on how to manage worsening symptoms such as exacerbations, and when it is appropriate to call for an ambulance. Using LINQ provides a way of assessing the effectiveness of an educational process as reported by changes in the patients' needs and should be considered as a standard outcome measure to assess the educational components of PR programmes.

Table 4 Mean score of SWT, HADS, CRQ-SR, CCQ, SBPQ and SGRQ pre and post PR

	Pre PR	Post PR	Significance	Effect size
	Mean (SD)	Mean (SD)	<i>t</i>	<i>r</i>
SWT (<i>N</i> = 115)	266.9 (199.4)	374.6 (333.8)	-4.32**	0.38
HADS (<i>N</i> = 112)				
Anxiety	7.3 (4.3)	5.9 (3.7)	4.48**	0.39
Depression	6.1 (3.7)	4.9 (3.3)	4.89**	0.42
CRQ-SR (<i>N</i> = 47)				
Total score	75.2 (19.9)	91.2 (17.0)	-6.44**	0.70
Dyspnoea	13.6 (6.3)	17.2 (6.0)	-4.06**	0.52
Fatigue	14.2 (4.0)	17.3 (4.5)	-5.28**	0.61
Emotional Function	30.5 (9.3)	35.2 (7.5)	-4.71**	0.57
Mastery	17.3 (5.8)	21.1 (4.7)	-5.74**	0.65
CCQ (<i>N</i> = 10)				
Total score	2.3 (1.1)	2.0 (0.8)	0.98	ns
Symptom	2.6 (1.5)	2.6 (1.0)	0.00	ns
Functional	2.1 (1.4)	1.5 (1.2)	1.86	ns
Mental State	1.8 (0.9)	1.5 (1.3)	0.72	ns
SBPQ total (<i>N</i> = 41)	11.9 (5.7)	10.7 (5.0)	1.83	ns
SGRQ (<i>N</i> = 46)				
Total score	53.0 (14.7)	48.1 (13.0)	2.87*	0.35
Symptom	59.7 (19.6)	55.0 (18.2)	1.78	ns
Activity	71.9 (18.3)	71.8 (17.2)	0.07	ns
Impact	40.1 (18.1)	32.7 (14.7)	3.46**	0.34

Note: * $p < 0.01$; ** $p < .001$. Effect sizes (*r*): small (0.10), moderate (0.30), and large (0.50). ns: indicates non significant changes, therefore effect size calculations were not applicable.

In this study, patients undergoing PR showed clinically important improvements in the SWT, HADS and CRQ-SR scores. For instance, the minimum clinically important difference in SWT is 48 m, and the mean improvement in our study was over 100 m. These results indicate that the study sites used in this study deliver effective PR, confirming their value as a benchmark for newer programmes. Baseline scores on the LINQ were unrelated to measures of disease severity or psychological distress suggesting that information needs is a different kind of measure to that normally considered in outcome research. We found only one significant positive correlation between change in LINQ and change in other variables, but this may have occurred due to chance as a result of multiple correlation testing.

Although change score are problematic due to lack of reliability, these results could have occurred by chance (due to multiple testing) and do not provide any clear evidence that change in LINQ is associated with positive change in other outcome measures.

Education is important for patients so that they can understand their disease and develop healthy behaviour patterns that can improve their healthcare utilisation and reduce their disability.⁴⁻⁶ To change behaviour, patients need more than factual information, they need to have their concerns addressed, such as the fear that exercise may be harmful or uncomfortable, and they need to understand the need to change their behaviour.¹⁶ At present it is clear that patients with COPD have a poor understanding of their disease and how to manage it.^{10,17}

Further studies are needed to examine the effect of education programmes, including PR, on changing behaviour patterns in the long term. To develop optimal education systems, the impact of the programme should be measured on key components such as information needs and adoption of healthy behaviour, as well as outcomes such as health status and exercise capacity. Use of the LINQ would allow comparison of different education methods in improving patients' information needs.

One limitation of this study is that the PR programmes differed substantially in terms of the number of sessions per week and the variety of staff involved at each site. However, similar improvements were seen in all the programmes. Published evidence suggest that PR programmes should be twice weekly,⁵ but no differences were seen between the LINQ and other outcomes across the sites. This was not a primary end-point and further studies are needed to assess the optimum structure of rehabilitation programmes. A further limitation is that 32 patients did not complete the PR programmes. Potentially this could introduce a bias towards the change in information needs over time, although the subjects' baseline data did not differ significantly from those participants that completed PR.

This study shows the LINQ is a practical tool for detecting areas where patients need education and is sensitive to change after PR, since all domains improved significantly except smoking. The quality of the education component of PR can be assessed using the LINQ which should be considered as a routinely collected outcome measure. However, we did not find a relationship between change in educational need and change in other outcome measures. Further research is needed to understand the

relationship between education during PR, change in information needs and change in quality of life and exercise tolerance.

Acknowledgements

Mrs G. Ackers — East Devon Pulmonary Rehabilitation Programme
 Mrs F. Dyer — Respiratory Care Team, Chertsey, Surrey Primary Care Trust
 Dr C. Bolton, Dr T. Griffiths & Mrs E. Hilsden — Llandough Hospital, Cardiff
 Dr K. Gruffydd-Jones — Box Surgery, Wiltshire
 Mrs B. Harmon, Mrs J. Bray, & Mrs J. Wells — Plymouth Pulmonary Rehabilitation Programme
 Mrs C. Langley — Chippenham Community Hospital
 Mrs L. Moore — Kings College Hospital, Southwark Primary Care Trust & Lambeth Primary Care Trust
 Dr B. Shackell — Respiratory Research Unit, Peninsula Medical School, Devon

Other interests

Dr Rupert Jones is a holder of a Researcher Development Award, Department of Health.

References

1. Global Initiative for Chronic Obstructive Lung Disease. Global strategy for the diagnosis, management and prevention of COPD. 2006.
2. Seemungal TA, Donaldson GC, Paul EA, Bestall JC, Jeffries DJ, Wedzicha JA. Effect of exacerbation on quality of life in patients with chronic obstructive pulmonary disease. *Am J Respir Crit Care Med* 1998 May;157(5 Pt 1):1418-22.
3. Wilkinson TM, Donaldson GC, Hurst JR, Seemungal TA, Wedzicha JA. Early therapy improves outcomes of exacerbations of chronic obstructive pulmonary disease. *Am J Respir Crit Care Med* 2004 Jun 15;169(12):1298-303.
4. Bourbeau J, Nault D, Dang-Tan T. Self-management and behaviour modification in COPD. *Patient Educ Couns* 2004; 52(3):271-7.
5. Nicl L, Donner C, Wouters E, et al. American Thoracic Society/European Respiratory Society statement on pulmonary rehabilitation. *Am J Respir Crit Care Med* 2006 Jun 15;173(12):1390-413.
6. National Institute for Clinical Excellence. *CG12 chronic obstructive pulmonary disease clinical guidelines*. London: Abba Litho Ltd.; 2004 Feb.
7. White R, Walker P, Roberts S, Kalisky S, White P. Bristol COPD Knowledge Questionnaire (BCKQ): testing what we teach patients about COPD. *Chron Respir Dis* 2007;3:123-31.
8. Hopp JW, Lee JW, Hills R. Development and validation of a pulmonary rehabilitation knowledge test. *J Cardiopulm Rehabil* 1989;7:273-8.
9. Hyland ME, Jones RCM, Hanney KE. The lung information needs questionnaire: development, preliminary validation and findings. *Respir Med* 2006;100:1807-16.
10. Zigmund AS, Snaith RP. The hospital anxiety and depression scale. *Acta Psychiatr Scand* 1983;67(6):361-70.
11. Singh SJ, Morgan MD, Scott S, Walters D, Hardman AE. Development of a shuttle walking test of disability in patients with chronic airways obstruction. *Thorax* 1992;47(12):1019-24.

13. Williams JE, Singh SJ, Sewell L, Guyatt GH, Morgan MD. Development of a self-reported Chronic Respiratory Questionnaire (CRQ-SR). *Thorax* 2001;56:954–9.
14. van der Molen T, Willemse BW, Schokker S, ten Hacken NH, Postma DS, Juniper EF. Development, validity and responsiveness of the Clinical COPD Questionnaire. *Health Qual Life Outcomes* 2003;1(1):13.
15. Jones PW, Quirk FII, Baveystock CM, Littlejohns PA. A self-complete measure of health status for chronic airflow limitation. *Am Rev Respir Dis* 1992;145:1321–7.
16. Horne R, Weinman J. Patients' beliefs about prescribed medicines and their role in adherence to treatment in chronic physical illness. *J.Psychosom Res.* 1999;47(6):555–67.
17. British Lung Foundation. Lost in translation, bridging the communication gap in COPD, http://www.lunguk.org/documents/lost_in_translation2.pdf [Accessed 11.12.07].
18. Cohen J. *Statistical power analysis for the behavioural sciences*. 2nd ed. New York: Academic Press; 1988.

4.1.4 Study Limitations

4.1.4.1 Study Design

Cohort studies are widely used within clinical and psychological research. They provide a methodology for examining a group of individuals with a common characteristic (referral for PR in this instance) against a specific research question (the ability of the LINQ to assess information needs pre- and post- intervention). Additionally, they allowed individuals from across multiple sites to be treated as a single group, thus strengthening the statistical power of the analysis. In addition to answering the primary research question, the cohort design also allows other outcomes to be evaluated.

Although the current studies looked for a change over time, the duration of the intervention was such that we cannot claim that these changes are longitudinal or permanent. A study design that might have helped answer the question about information needs over time and the long term usefulness of both the LINQ and PR would be a cross-sectional methodology. Cross-sectional studies have several characteristics that make them attractive to researchers. They are relatively inexpensive, quick and easy to do, are useful for generating and clarifying hypotheses, piloting new measures or technology and can lay the groundwork for decisions about future follow-up studies (Kraemer, 1994). They provide information about group differences or inter-individual differences (Miller, 2007).

Cross-sectional studies do not, however, provide information about changes or inter-individual differences in intra-individual change (Miller, 2007; Wohlwill, 1973). Cross-sectional studies are also subject to methodological concerns and limitations as discussed in section 3.2.4.1.

4.1.5 Contribution to knowledge and autoethnographic issues

4.1.5.1 Contribution to knowledge

The LINQ was the first tool designed with patients to assess their PR information needs rather than their level of knowledge. The LINQ allowed the clinical teams to tailor information provision to participants during PR, and also during routine clinical visits. The LINQ was subsequently identified by the British Thoracic Society PR guidelines as one of the tools that should be used to ensure the educational content of clinic visits is appropriate and improved/reviewed regularly.

The LINQ allows clinicians to identify individuals who need particular attention to their understanding of their disease, and specific areas of needs. It further showed that there was not a clear causal link between changes in educational need and outcomes over a PR intervention. It is also short and quick to complete, allowing it to be used in routine clinical practice.

Appendix 7 has an overview of how the research undertaken in paper 3 was disseminated prior to the paper's publication. It also provides a list of papers where paper 3 has been cited, and this is followed by a quality appraisal of the paper.

4.1.5.2 Autoethnographic issues

This was the first project where I was invited to help out, where my skills were requested. As such, I was able to seek out new experiences and understand more of the pragmatic aspects of setting up and running groups for and with people with different needs.

It was attending one of the PR groups organised by a NHS chest service, where it became clear to me that sometimes the people organising these groups do not always fully account for the people attending. For example, at one site the PR was booked to run on the first floor of a community clinic, where the lift had been out of service for more than 2 years. This meant that people with significant lung diseases and often highly reduced mobility had to walk up two flights of stairs simply to access the group. On more than one occasion, I stood at the bottom of the stairs half an hour before the group started and walked some of the attendees up, providing them an arm, and someone to talk to or just sit with when they need to sit down between flights of stairs. At another site, a group was arranged in a church hall, and a patient felt they could not attend as they were not Christian, but did not want to voice this to the staff.

These experiences meant I became explicitly aware of the needs of the individuals. Up to this point I had undertaken formal risk assessments of research environments; was there a fire escape, how many people could the room hold, had all the equipment been portable appliance tested etc. But I had always thought that participants' individual needs and requirements would have been taken in to consideration, in the same way as asking about allergies if we were organising an event where food was provided. So how did this change me? I ensured I fed back to the organisers of the events when I saw, or was made aware of any challenges being

experienced. In doing this I found out that although numerous people commented on a problem to me, or between themselves expressed a negative opinion, none of them had commented to the PR staff in person or on the evaluation sheet, even though there was specifically identified space to comment on the room/setting. Consequently, the organisers were often surprised when I told them, but in the majority of cases were able to arrange new venues where needed, and amend patient facing documents to obtain additional information, such as ethnicity and religion to ensure venues were acceptable.

I now endeavour to seek participants' opinions, and gain their views of the construction of an intervention, the venues where events will take place, and any aspects that they feel are important. At the end of the research or intervention, I specifically ask participants for their views around the practicalities of the event, couching the discussion in terms of patient involvement and the improvement of the service.

4.1.7 Future research questions

During the research, I noticed that there appeared to be a greater level of physical and psychological improvement in the patients whose partners/spouses attended with them. I discussed my thoughts with the team and a number of them felt the same. However, data had not been collected on these 'co-attendees', and no record has been made of those patients whose partners had come with them. So, the question I was left with was; is pulmonary rehabilitation more effective for people with COPD if their spouses participate in the programme as well?

Unfortunately, at the end of the project I was coming to the end of my contract and I did not have the opportunity to investigate this. In the years since the publication of the research in 2008, others have investigated this area and indicated that my thoughts and feelings were accurate (Chen et al 2017; Figueiredo et al 2016; Jeong & Yoo, 2015). This has gone on to show how our health and the interventions that we design are more than the clinicians, more than the patients' biology, more than their psychology and more than their social support. Although our treatments largely focus on one of these, we need to be aware of the larger systems that we all inhabit and how they impact on those we care for at home and at work.

4.2 Paper 4 - Clinical experience leading to the research work and production of the paper

I started working at the Diving Disease Research Centre (DDRC; now DDRC Healthcare) in July of 1999. I approached DDRC with a project idea, and although generally supportive they asked me to develop the idea, and learn to dive. I did these two things and returned with some new skills and my first research proposal. While seeking funding, I was trained as a Hyperbaric chamber attendant and operator, and as a Diver Medic Technician. This was an exciting time and I learnt a lot, but I was not actively using my Psychology knowledge and skills. It also became clear that this 'Research Centre' was not actually investigating its impact on the patients it was treating. It was from this point that DDRC encouraged me to undertake research into the impact of HBOT, on the QoL of patients who had undergone radiotherapy for the treatment of head and neck cancer (HNC).

The Undersea and Hyperbaric Medical Society defines HBOT as an intervention in which an individual breathes near 100% oxygen intermittently while inside a hyperbaric chamber that is pressurised to greater than sea level pressure (1 atmosphere absolute, or ATA). For clinical purposes, the pressure must equal or exceed 1.4 ATA while breathing near 100% oxygen. Current evidence supports its use in 14 medical conditions, one of which is delayed radiation tissue damage, which is an umbrella term covering soft tissue and osteoradionecrosis. The provision of HBOT is very specialised and DDRC is one of only six category one chambers within the United Kingdom enabled to provide this therapy on a 24 hour routine or emergency basis.

4.2.1 Research Questions

The work reported in paper 4 investigated how receiving HBOT before and after treatment for the long term side effects, of treatment for HNC (requirement to have teeth removed, dental implant placed, or to treat osteoradionecrosis) affected the QoL of these patients.

4.2.2 Study Design

In wanting to understand the impact of a treatment on a cohort of people, it was decided that a pre-, post-cohort design was most suitable. DDRC is a charity which provides HBOT as specialist treatment to NHS patients. Even more than within the

NHS, the patients referred are a limited subset of people who have been treated for HNC. In order to be referred, at least part of their treatment had to have been radiotherapy. The medically induced problem they are being managed for are either due to the toxic effect of radiotherapy or the heightened risk of damage occurring due to surgical intervention within the irradiated field.

Radiation injuries can be sub-classified as acute, sub-acute or delayed complications (Rubin & Cassarrett, 1968). Acute injuries are due to direct and near immediate cellular toxicity caused by free radical-mediated damage to cellular DNA. This type of radiation injury is usually short term and treated symptomatically, for example treatments for oral thrush, and skin burns. However, they can be very debilitating during their duration. Sub-acute injuries typically onset two to three months after treatment and may persist for several months. Delayed radiation complications are typically seen after a period of six months or more and may develop many years after the radiation exposure. Sometimes, acute injuries are so severe that they never resolve and evolve to become chronic injuries indistinguishable from delayed radiation injuries (Dorr & Hendry, 2001). Often, delayed injuries are precipitated by another incident or intervention such as surgery within the radiation field.

It has been suggested that the impact of hyperbaric oxygen in terms of its beneficial effects in irradiated tissues includes: 1) Stimulation of angiogenesis and secondarily improvement of tissue oxygenation; 2) Reduction of fibrosis; and 3) Mobilisation and stimulation of stem cells within irradiated tissues (Marx, 1999; Trott, 1984; Goldstein, Gallaher, Bauer et al, 2006).

The great majority of the research has focused on these biological effects; evaluation of the impact of QoL related to HBOT was very limited. General QoL research with people treated for HNC has been ongoing for over 30 years, and the questionnaires used in these studies were reviewed, and guided the setting of research objectives and selection of the research measures. We recruited consecutive prospective patients who had been referred for HBOT for delayed radiation tissue damage where surgery was planned over a period of more than four years.

4.2.3 Impact of perioperative hyperbaric oxygen therapy on the quality of life of maxillofacial patients who undergo surgery in irradiated fields

Clinical Paper
Head and Neck Oncology

Impact of perioperative hyperbaric oxygen therapy on the quality of life of maxillofacial patients who undergo surgery in irradiated fields[☆]

S. A. Harding¹, S. C. Hodder²,
D. J. Courtney³, P. J. Bryson¹

¹Hyperbaric Medical Centre, Derriford, Plymouth, Devon, UK; ²Department of Oral & Maxillofacial Surgery, Morriston Hospital, Swansea, UK; ³Department of Oral & Maxillofacial Surgery, Derriford Hospital, Plymouth, Devon, UK

S. A. Harding, S. C. Hodder, D. J. Courtney, P. J. Bryson: Impact of perioperative hyperbaric oxygen therapy on the quality of life of maxillofacial patients who undergo surgery in irradiated fields. *Int. J. Oral Maxillofac. Surg.* 2008; xxx: xxx-xxx. Crown Copyright © 2008 Published by Elsevier Ltd on behalf of International Association of Oral and Maxillofacial Surgeons. All rights reserved.

Abstract. From 2001 to 2005, 66 patients referred for perioperative hyperbaric oxygen therapy (HBO₂) for debridement of necrotic tissue or prevention of radionecrosis were assessed with quality of life measures, before and after completion of HBO₂ and surgery. The Medical Outcomes Short Form 36 (SF-36) and Hospital Anxiety and Depression Scale (HADS) showed no significant changes. The European Organisation for Research and Treatment of Cancer Core (EORTC-C30) questionnaire showed significant improvement in pain, global health, and dyspnoea ($p = 0.011$; $p = 0.027$; $p = 0.008$, respectively). The Head and Neck sub-module (H&N35) identified significant improvements in teeth, dry mouth and social contact ($p = 0.002$; $p = 0.038$; $p = 0.029$, respectively). The University of Washington Scale (UW), showed significant changes in relation to chewing and shoulders ($p = 0.031$; $p = 0.047$). When sub-group analysis using 'osteoradionecrosis' and 'dental extraction or implants' was performed on the EORTC and UW data, variations in the patterns of significance were found. Adjunctive HBO₂ should be considered for the treatment and prevention of some of the long-term complications of radiotherapy.

Keywords: hyperbaric oxygenation; quality of life; radiotherapy.

Accepted for publication 10 April 2008

In 2005, 4559 people were newly registered as having cancer in the head and

neck region (ICD-00 code; C00-C14) in the UK²¹. Radiotherapy is widely used in the treatment of a range of primary and metastatic neoplasms in the head and neck region. Between 60% and 80% of all people with head and neck cancers, and

nearly 100% of people with T3/4 staged disease in the UK, receive radiotherapy either as their primary therapy or as an adjunct to surgery and chemotherapy¹³. Treatments are carefully planned to minimise exposure of surrounding normal tis-

[☆] Undersea and Hyperbaric Medical Society 37th & 38th Annual Scientific Meeting.

sues to ionizing radiation, but, there is inevitably some transient or permanent tissue damage to the surrounding structures. The resulting complications are often associated with swallowing, taste, chewing, sensory and motor function and pain, and adversely affect the patients' quality of life (QoL). Studies suggest that short-term morbidity is generally stable after as little as 1 year, but that it can take up to 3 years for some QoL measures to return to near pre-cancer levels⁴. Despite this, some factors, including fibrosis, sensation of taste and smell, dry mouth, sticky saliva and sexuality, are particularly resistant to these gradual improvements. There is anecdotal evidence that hyperbaric oxygen therapy (HBO₂) may improve some of these issues.

Surgical intervention in a heavily irradiated field may result in delayed wound healing, dehiscence or infection^{12,22}. HBO₂ is widely proposed to reduce associated risks^{7,9,11,15,19} although a review, retrospective analysis and a recent study did not support its use^{1,14,25}. Recently, treatment combining pentoxifylline and tocopherol has been found to be of use in the treatment of radiation-induced fibrosis and may prove beneficial in the treatment of radiation tissue damage⁶. The use of HBO₂ in this therapeutic area remains a topic of debate. Comprehensive literature reviews in 2002 and 2004^{9,10} indicated that HBO₂ is an effective treatment for established radiation tissue damage at multiple anatomical sites. The research of Marx and others^{9-11,17,18} supports the theory that HBO₂ acts as an adjunct to healing in irradiated tissue by stimulating angiogenesis and fibroplasia. Data from a randomised clinical trial¹⁸ supports the use of HBO₂ as a prophylactic measure when tooth removal is performed in an irradiated area. The use of HBO₂ in the placement of osseointegrated dental implants in an irradiated area is supported by numerous studies, which have recently been comprehensively reviewed by Granström¹¹. These findings have culminated in patients receiving HBO₂ pre and post (peri) operatively in many centres.

There is a growing body of work that assesses the impact of treatment for head and neck cancers and several questionnaires and surveys have been developed to assess the QoL in this patient group^{2,3,16,24,26,27}. No significant research has been conducted to investigate the changes in QoL in these patients as a result of adjunctive HBO₂. The authors conducted a cohort study of patients who received adjunctive HBO₂ peri-operatively to treat the complications of prior

radiotherapy to establish the impact of this treatment on QoL.

Materials and methods

Ethics

Ethical approval was granted from the Local Research Ethics Committee following British Psychological Society Guidelines and the Declaration of Helsinki. The study was explained via an information sheet and questions were answered. Written consent was obtained.

Participants

From 2001 to 2005, 66 patients (48 males and 18 females; mean age 56.6 years) referred consecutively for HBO₂ following radiotherapy to head and neck cancers were recruited to complete questionnaires before commencing their HBO₂ therapy and then again on completion of their therapy. Thirty-eight volunteers were referred for treatment to cover dental extraction or intraoral implant placement, and 28 were having treatment for 'osteoradionecrosis'.

Inclusion and exclusion criteria

All patients were over the age of 18 years and spoke English as their first language.

None of the patients had previously undergone HBO₂. Patients were referred for peri-operative HBO₂ for either debridement of necrotic tissue, or prevention of radionecrosis due to dental extraction or for intraoral implant placement within an irradiated field. All referring consultants were asked for additional information or comments regarding patient inclusion or exclusion from the trial prior to attendance at the hyperbaric unit.

HBO₂ regimen

Completing patients received between 14 and 40 (mean = 26.4, SD = 4.3) therapies prior to surgical interventions, and between 6 and 23 (mean = 16.3, SD = 3.5) therapies following surgery, in a multiplace chamber at the Hyperbaric Medical Centre in Plymouth, UK. All were treated at 2.2 ATA (12 m) for a total of 90 minutes breathing 100% oxygen, which was administered in 2 oxygen breathing periods of 45 minute each, separated by a 5 minute air break (Fig. 1). Treatments were conducted on 5 working days each week. Oxygen was delivered via an Amron Oxygen Treatment Hood, or a Sea-Long Series 7000 Mask.

Questionnaires

There are no QoL questionnaires designed to be used specifically in the field of hyperbaric medicine. The measures used in this research have been developed and validated in settings such as outpatient clinics and in the hospital environment, and were deemed appropriate for the assessment of change in this study.

The Hospital Anxiety and Depression Scale (HADS)³⁰ questionnaire is a self-administered scale composed of statements relevant to either generalized 'anxiety' or 'depression', the latter being largely (but not entirely) composed of reflections of the state of anhedonia (the inability to gain pleasure from normally pleasurable experiences). Each item has a 4-point (0-3) response category with possible scores ranging from 0 to 21 for anxiety and 0 to 21 for depression. A score of 0 to 7 on either scale indicates the respondent falls within 'normal' ranges for anxiety and depression; 8-11 is mild; 12-15 is moderate; and 16-21 is a severe level of either trait.

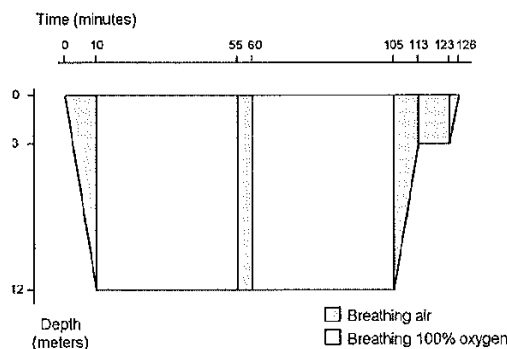


Fig. 1. Treatment profile.

The Medical Outcomes Short Form 36 (SF-36)²⁸ questionnaire is a self-administered instrument constructed to represent 8 of the most important health concepts: physical functioning, role-physical, bodily pain, general health, vitality, social functioning, role-emotional and mental health. Each question has between 3 and 6 response options using a Likert-type scale. The SF-36 is referred to as a generic measure as it assesses health concepts that are relevant to everyone's functional status and well-being²⁷. Such measures are termed generic, and are universally valued because they are not age, disease or treatment specific. Generic health measures assess health-related QoL outcomes, namely those known to be most directly affected by disease and treatment.

The University of Washington Quality of Life Version 4 (UW-QoL)²⁴ questionnaire is a self-administered instrument consisting of 15 questions; 12 are disease-specific items (pain, appearance, activity, recreation, swallowing, chewing, speech, shoulder, taste, saliva, mood and anxiety), and 3 are general questions. The general questions were not considered in this study because they relate directly to the participants' experience of cancer and changes over a shorter period of time (7 days) than that covered by the study. Each of the 12 included questions has 3-6 response options using a Likert-type scale. Each item is scored from 0 to 100, with higher scores indicating better QoL. This results in a summary score of 0-1200 for disease-specific items.

The European Organization for Research and Treatment of Cancer (EORTC)⁸ is a modular instrument designed to bridge the roles of disease-specific and global QoL scales. It is a patient-based, self-administered multidimensional instrument. Version 3.0 of the EORTC QLQ-C30 core questionnaire consists of 30 questions organised into 5 domains: physical functioning, role functioning, cognitive functioning, emotional functioning, and social functioning; 3 symptom scales: fatigue, pain, nausea and vomiting; a global scale (Global Health Status/QoL), and 6 single items (dyspnoea, appetite loss, insomnia, constipation, diarrhoea, and financial difficulties). The EORTC Head and Neck (QLQ-H&N35)² questionnaire, consists of 35 questions organised into 7 domains: pain, swallowing, senses problems, speech problems, trouble with social eating, trouble with social contact, and less sexuality; as well as 11 single items (teeth, opening mouth, dry mouth, sticky saliva, coughing,

felt ill, pain killers, nutritional supplements, feeding tube, weight loss, weight gain).

The participants completed the questionnaires unsupervised.

Analysis

Analysis was conducted on the responses from the 66 (48 males and 18 females; mean age 56.6 years) participants who completed both pre- and post-questionnaires, using the *t*-test in SPSS (Version 15.0). Assumptions of normality were tested and found to be valid. Stepwise regression was used to assess the relationship between QoL measures and the length of time from treatment for cancer to HBO₂.

Results

All participants completed the questionnaire battery on both occasions. Participants were on average 6 years 5 months (min 5 months; max 27 years 8 months; St Dev, 5.6) post cancer diagnosis. The regression analysis yielded no association with QoL outcome and time from cancer.

Analysis using HADS revealed that there were no significant differences in anxiety or depression (Table 1). The mean values for both sub-scales at both time points fall within 'normal' as defined by the questionnaire (0-7). The range of scores from HADS post HBO₂ was 0-15 on the anxiety sub-scale and 0-13 on the depression sub-scale. When the whole dataset was split into the 4 classifications outlined in HADS, it was found that 13% of participants reached moderate levels of anxiety (none reached severe) and 2% reached the moderate categories on depression (none reached severe).

Using the SF-36 no significant improvements were found (Table 2).

The UW-QoL Scale indicated a significant decline in chewing for the participants as a whole for the duration of this

study. The extraction or implants sub-group also showed significant improvement in relation to 'shoulder' with a reduction in 'chewing' (Table 3).

Changes in QoL were evident using the EORTC QLQ-C30 (Table 4) where all patients showed significant improvement in 'Global Health Status/QoL', 'pain' and 'dyspnoea'. The improvements in 'pain' were also evident in the extraction or implants sub-group.

The EORTC QLQ-H&N35 identified significant ameliorations in relation to 'teeth' and 'dry mouth'. The differences with 'teeth' were also manifest in the extraction or implants sub-group. The debriement group showed significant improvements in 'opening mouth' and 'pain killers'. This questionnaire also revealed some declines in QoL. 'Social contact' declined significantly in the whole data set and in the extraction or implants sub-group. Worsening of sexuality was evident in the debriement sub-group (Table 5).

Not all participants completed all questions on all questionnaires at both time points. The most noticeable example was on HADS; the number of people completing this questionnaire to a degree that allows statistical analysis was 56 on both subscales. In most cases where the questionnaire was not fully completed only one question was not answered (although it was not always the same question). No consistent reason was given for non-completion, although one participant 'did not like' the questionnaire so completed the others and not HADS. The non-completion of questions only occurred to the same extent in one sub-scale of one other questionnaire. Fifty-nine people responded to the questions regarding sexuality on the EORTC QLQ-H&N35. These patients tended to indicate that they were either not married or widowed, and on occasion noting that they no longer 'wanted' or 'were able to have' sex. A similar response rate has been experienced using these two questionnaires in

Table 1. Statistics for Hospital Anxiety and Depression Scale (HADS) scores

HADS	N	t	DF	T ₁	T ₂	T ₁ -T ₂	p
Anxiety - All	56	0.073	55	5.96	5.93	0.03	0.942
Extraction or implants	30	-0.086	29	6.83	6.90	0.07	0.932
'osteoradionecrosis'	25	0.270	24	4.92	4.76	-0.16	0.790
Depression - All	56	-1.153	55	4.36	4.80	0.44	0.254
Extraction or implants	31	-1.270	30	4.35	5.03	0.68	0.790
'osteoradionecrosis'	25	-0.282	24	4.36	4.52	0.16	0.780

N is the number of participants data used in the calculation, t is the critical value used in combination with degrees of freedom to assess significance, DF is degrees of freedom, which is the number of scores that are free to vary in calculating the statistic, p is the level of significance, T₁ = time point 1, T₂ = time point 2. A lower score represents a reduced level of anxiety and depression.

Table 2. Statistics for Medical Outcomes Short Form 36 (SF-36) scores

SF-36	N	t	DF	T ₁	T ₂	T ₁ -T ₂	p
Physical functioning – All	65	0.623	64	44.89	44.47	-0.42	0.535
Extraction or implants	37	-0.143	36	44.41	44.52	1.11	0.887
'osteoradionecrosis'	28	0.965	27	45.53	44.41	-1.12	0.343
Role-physical – All	62	-0.765	61	35.13	36.23	1.10	0.447
Extraction or implants	36	-0.069	35	37.19	37.33	0.14	0.946
'osteoradionecrosis'	26	-1.164	25	32.27	34.72	2.45	0.256
Bodily pain – All	65	-1.143	64	42.76	44.21	1.45	0.257
Extraction or implants	37	-1.493	36	43.52	45.69	2.17	0.144
'osteoradionecrosis'	28	-0.226	27	41.74	42.26	0.52	0.823
General health – All	66	-1.150	65	43.25	44.30	1.05	0.254
Extraction or implants	38	-1.118	37	40.90	42.13	1.23	0.271
'osteoradionecrosis'	28	-0.511	27	46.43	47.23	0.80	0.613
Vitality – All	65	-0.480	64	47.29	47.86	0.57	0.633
Extraction or implants	38	-0.488	37	46.42	47.16	0.74	0.628
'osteoradionecrosis'	27	-0.174	26	48.51	48.85	0.34	0.863
Social functioning – All	63	0.327	62	42.91	42.48	-0.43	0.745
Extraction or implants	37	-0.442	36	42.70	43.44	0.74	0.661
'osteoradionecrosis'	26	0.970	25	43.21	41.12	-2.09	0.341
Role-emotional – All	64	-0.307	63	33.59	34.13	0.54	0.760
Extraction or implants	38	-0.455	37	35.52	36.44	0.92	0.652
'osteoradionecrosis'	26	0.000	25	30.76	30.76	0.00	1.000
Mental health – All	64	-1.320	63	48.29	49.83	1.54	0.192
Extraction or implants	38	-0.958	37	46.60	47.93	1.33	0.344
'osteoradionecrosis'	26	-0.894	25	50.77	52.61	1.84	0.380

N is the number of participants data used in the calculation, t is the critical value used in combination with degrees of freedom to assess significance, DF is degrees of freedom, which is the number of scores that are free to vary in calculating the statistic. p is the level of significance. T₁ = time point 1, T₂ = time point 2. A higher score indicates better quality of life.

other studies of similar patient group (S. Rogers, pers. comm.).

Of the 66 participants in this study 6 (9%) had been discharged from hospital follow up or their referring consultants could not supply information. The data for the remaining 60 is of varied quality, but can be broadly categorised into; healed, ongoing problems, improved and healing slowly, occasionally shedding sequestra, or cancer recurrence/died (Table 6). The Hyperbaric Medical Centre in Plymouth is a registered charity and as such is outside the National Health Service. This can lead to difficulties in obtaining complete and detailed follow-up information on all the patients treated.

Discussion

QoL measures have been widely used in the assessment of patients with head and neck malignancy. They are a valuable tool since these cancers, and the treatment that patients receive, can have a significant impact on individuals' QoL. There are no data regarding improvements in QoL in this patient group following treatment with adjunctive HBO₂.

Previous QoL studies involving head and neck cancer patients who did not receive HBO₂ have recruited about 100 patients^{5,23,29}. A formal power calculation was not performed since there is no previous data in this field involving HBO₂.

In the light of these findings the authors could take the research further by constructing a control group. Control groups in HBO₂ studies are often controversial and numerous strategies have been used. These include blinded treatments, sham treatments and the use of normal or rarefied air (to simulate surface oxygen partial pressure) at typical HBO₂ treatment pressures. These control group strategies require significant operational and financial commitment. Owing to the nature and workload of the authors' unit, the inclusion of a control group was not feasible. With this in mind a repeated measures design was chosen for the study, providing some degree of internal control to the data collection. Multi-centre studies could address these problems and allow for the addition of functional measures.

The authors feel that the improvements in QoL described may be attributed to the combination of HBO₂ and surgery. After the initial cancer treatment there is often a high level of depressive symptomatology that impairs QoL⁴. Although many facets of QoL approach normal, some factors, including sensation of taste and smell, dry mouth, and sticky saliva are resistant to improvement. In this study, the mechanism of the referral and treatment process prevented the authors from assessing the stability of QoL measures in their patient group; however the mean average of the patients assessed was 6 years post diag-

nosis and subsequent treatment. Given the suggestion by CHANDU et al.⁴ that short-term morbidity can be generally stable after 1 year, and nearing pre-cancer levels by 3 years, the authors think that it is reasonable to assume QoL in these patients to be stable prior to HBO₂ and surgery.

HADS has been shown²⁰ to have good psychometric properties in terms of factor structure, sub-scale inter-correlation, homogeneity and internal consistency. The properties of HADS have been found to be robust across a wide spectrum of sub-samples, including groups with somatic problems, mental problems and different strata defined by age, education and gender. This questionnaire failed to demonstrate any significant differences in the present study, however a few participants fell outside the 'normal' ranges of the questionnaire.

The SF-36 is often used in medical trials as a stand-alone measure and has been shown to be both reliable and valid in a clinical setting. Although not used in a hyperbaric study with this patient group previously, there is no evidence to suggest that it is not appropriate for these purposes. The lack of significant changes in this study using the SF-36 may be due to the global nature of the questionnaire and that the changes experienced by the participants are attributable to the combination of surgery and HBO₂.

Table 3. Statistics for University of Washington Quality of Life Version 4 (UW-QoL) scores

UoW	N	t	DF	T ₁	T ₂	T ₁ -T ₂	p
UW 1 – Pain – All	65	-1.540	64	60.00	64.62	4.62	0.128
Extraction or implants	37	-1.348	36	61.49	66.89	5.40	0.186
'osteoradionecrosis'	28	-0.779	27	58.04	61.61	3.57	0.443
UW 2 – Appearance – All	65	0.252	64	76.92	76.15	-0.77	0.802
Extraction or implants	37	0.320	36	78.38	77.03	-1.35	0.750
'osteoradionecrosis'	28	0.000	27	75.00	75.00	0.00	1.000
UW 3 – Activity – All	65	-0.780	64	67.23	68.85	1.62	0.438
Extraction or implants	37	-2.021	36	66.22	70.95	4.73	0.051
'osteoradionecrosis'	28	0.697	27	68.57	66.07	2.50	0.492
UW 4 – Recreation – All	65	1.731	64	72.62	69.23	-3.39	0.088
Extraction or implants	37	0.522	36	73.51	72.30	-1.21	0.605
'osteoradionecrosis'	28	1.888	27	71.43	65.18	-6.25	0.070
UW 5 – Swallowing – All	65	0.863	64	53.08	46.92	-2.16	0.391
Extraction or implants	37	0.542	36	74.86	73.24	-1.62	0.591
'osteoradionecrosis'	28	0.665	27	74.29	71.43	-2.86	0.512
UW 6 – Chewing – All	65	2.049	64	53.08	46.92	-6.16	0.045
Extraction or implants	37	2.744	36	59.46	48.65	-10.81	0.009
'osteoradionecrosis'	28	0.000	27	44.64	44.64	0.00	1.000
UW 7 – Speech – All	65	-0.422	64	77.92	78.92	1.00	0.674
Extraction or implants	37	0.378	36	80.81	79.46	-1.35	0.708
'osteoradionecrosis'	28	-1.476	27	74.11	78.21	4.10	0.151
UW 8 – Shoulder – All	65	-0.952	64	70.31	72.85	2.54	0.345
Extraction or implants	37	-2.055	36	67.57	74.59	7.02	0.047
'osteoradionecrosis'	28	0.841	27	73.93	70.54	-3.39	0.408
UW 9 – Taste – All	65	-0.246	64	62.62	63.23	0.61	0.806
Extraction or implants	37	0.470	36	64.86	63.51	-1.35	0.642
'osteoradionecrosis'	28	-0.731	27	59.64	62.86	3.22	0.471
UW 10 – Saliva – All	65	-1.877	64	48.62	53.69	5.07	0.065
Extraction or implants	37	-1.154	36	45.95	50.27	4.32	0.256
'osteoradionecrosis'	28	-1.545	27	52.14	58.21	6.07	0.134
UW 11 – Mood – All	61	0.743	60	75.33	73.36	-1.97	0.460
Extraction or implants	33	0.170	32	72.58	71.97	-0.61	0.866
'osteoradionecrosis'	28	0.891	27	78.57	75.00	-3.57	0.381
UW 12 – Anxiety – All	60	0.747	59	73.33	71.33	-2.00	0.458
Extraction or implants	33	0.675	32	70.30	67.88	-2.42	0.866
'osteoradionecrosis'	27	0.362	26	77.04	75.56	-1.48	0.381
UW Total – All	60	-0.063	59	794.83	795.75	0.92	0.950
Extraction or implants	33	-0.327	32	788.18	794.39	6.21	0.745
'osteoradionecrosis'	27	0.243	26	802.96	797.41	-5.55	0.810

N is the number of participants data used in the calculation, t is the critical value used in combination with degrees of freedom to assess significance, DF is degrees of freedom, which is the number of scores that are free to vary in calculating the statistic. p is the level of significance. T₁ = time point 1, T₂ = time point 2. A higher score indicates better quality of life.

UW-QOL is a short instrument best administered, as it was here, alongside a general QoL measure. The reason for this is its clinical nature; by combining the UW-QOL scale with another, greater specificity and contextualisation can be achieved. This has supplied proven reliability, responsiveness and validity and has demonstrated itself to be a useful instrument for surgically treated head and neck cancer patients. This questionnaire is designed for use with people currently

Table 4. Statistics for European Organisation for Research and Treatment of Cancer Core (EORTC QLQ-C30) data

	EORTC QLQ-C30	N	t	DF	T ₁	T ₂	T ₁ -T ₂	p
5 Domains	Physical functioning* – All	64	1.088	63	80.00	78.75	-1.25	0.281
	Extraction or implants	36	0.644	35	79.81	79.07	-0.74	0.524
	'osteoradionecrosis'	28	0.869	27	80.24	78.33	-1.91	0.392
	Role functioning* – All	62	0.720	61	74.46	72.04	-2.42	0.474
	Extraction or implants	35	0.695	34	74.29	71.90	-2.38	0.492
	'osteoradionecrosis'	27	0.386	26	74.69	72.22	-2.47	0.703
	Cognitive functioning* – All	64	0.925	63	79.69	78.12	-1.56	0.359
	Extraction or implants	36	1.000	35	75.93	73.61	-2.32	0.324
	'osteoradionecrosis'	28	0.238	27	84.52	83.93	-0.59	0.813
	Emotional functioning* – All	63	-1.151	62	73.55	75.93	2.38	0.254
	Extraction or implants	36	-0.919	35	70.37	72.69	2.32	0.365
	'osteoradionecrosis'	27	-0.700	26	77.78	80.25	2.47	0.490
	Social functioning* – All	64	0.000	63	69.79	69.79	0.00	1.000
	Extraction or implants	36	0.117	35	71.90	71.43	-0.47	0.908
	'osteoradionecrosis'	28	0.000	27	67.33	67.33	0.00	1.000

Table 4 (Continued)

EORTC QLQ-C30		N	t	DF	T ₁	T ₂	T ₁ -T ₂	p
Symptoms Scales	Fatigue [†] – All	62	-1.240	61	30.82	34.23	3.41	0.220
	Extraction or implants	35	-0.976	34	33.33	36.19	2.86	0.336
	'osteoradionecrosis'	27	-0.806	26	27.57	31.69	4.12	0.428
	Pain [†] – All	63	2.633	62	37.83	29.89	-7.94	0.011
	Extraction or implants	35	2.130	34	36.67	30.48	-6.19	0.040
	'osteoradionecrosis'	28	1.753	27	39.29	29.17	-10.12	0.091
	Nausea and vomiting [†] – All	63	-0.159	62	9.25	9.52	0.26	0.874
	Extraction or implants	35	-0.215	34	10.48	10.95	0.47	0.831
	'osteoradionecrosis'	28	0.000	27	7.74	7.74	0.00	1.000
Global Scales	Global Health Status/ QoL* – All	63	-2.266	62	60.45	66.01	5.56	0.027
	Extraction or implants	35	-1.813	34	59.29	65.95	6.66	0.079
	'osteoradionecrosis'	28	-1.342	27	61.90	66.07	4.17	0.191
Single Item Scales	Dyspnoea[†] – All	64	2.722	63	15.63	7.29	-8.34	0.008
	Extraction or implants	36	1.848	35	15.74	8.33	-7.41	0.073
	'osteoradionecrosis'	28	1.982	27	15.48	5.95	-9.53	0.058
	Appetite loss[†] – All	64	0.129	63	23.96	23.44	-0.52	0.898
	Extraction or implants	36	-0.183	35	25.00	25.93	0.93	0.856
	'osteoradionecrosis'	28	0.359	27	22.61	20.24	-2.38	0.722
	Insomnia[†] – All	63	0.000	62	32.80	32.80	0.00	1.000
	Extraction or implants	36	-1.972	35	32.41	37.96	5.55	0.057
	'osteoradionecrosis'	27	1.363	26	33.33	25.93	-7.40	0.185
	Constipation[†] – All	64	0.869	63	16.15	13.54	-2.61	0.388
	Extraction or implants	36	-0.239	35	14.81	15.74	0.93	0.812
	'osteoradionecrosis'	28	1.536	27	17.86	10.71	-7.15	0.136
	Diarrhoea[†] – All	64	1.012	63	26.04	19.27	-6.77	0.315
	Extraction or implants	36	1.046	35	34.26	22.22	-12.04	0.303
	'osteoradionecrosis'	28	0.000	27	15.48	15.48	0.00	1.000
	Financial impact[†] – All	64	-0.841	63	32.29	35.93	3.65	0.404
	Extraction or implants	36	-1.540	35	27.78	35.19	7.41	0.132
	'osteoradionecrosis'	28	0.153	27	38.10	36.90	-1.20	0.879

N is the number of participants data used in the calculation, t is the critical value used in combination with degrees of freedom to assess significance, DF is degrees of freedom, which is the number of scores that are free to vary in calculating the statistic. p is the level of significance. T₁ = time point 1, T₂ = time point 2.

*Higher score indicates better function.

†Higher score indicates more symptoms.

experiencing head and neck cancer, therefore it may not be surprising that only one significant difference was found for the group as a whole (chewing) because they are on average 6 years post cancer treat-

ment. No changes were noted in the debilitation sub-group, but the extraction and implant sub-group showed the same decline in chewing as the group as a whole. This decline is possibly a result

of many of the patients undergoing dental extractions having no, or fewer, teeth after surgery or very recent implant placement. The reported improvement in 'shoulder' in the implants and extraction can not be

Table 5. Statistics for European Organization for Research and Treatment of Cancer, Head and Neck (EORTC QLQ-H&N35) data

EORTC QLQ-H&N35		N	t	DF	T ₁	T ₂	T ₁ -T ₂	p
7 Domains	Pain – All	62	1.267	61	35.89	31.99	-3.90	0.210
	Extraction or implants	34	0.637	33	33.58	31.13	-2.45	0.528
	'osteoradionecrosis'	28	1.127	27	38.69	33.04	-5.65	0.270
	Swallowing – All	63	0.281	62	21.30	20.77	-0.53	0.780
	Extraction or implants	35	0.107	34	21.67	21.43	-0.24	0.915
	'osteoradionecrosis'	28	0.275	27	20.83	19.94	-0.89	0.785
	Senses problems – All	62	0.508	61	28.23	26.61	-1.62	0.614
	Extraction or implants	35	0.122	34	27.62	27.14	-0.48	0.903
	'osteoradionecrosis'	27	0.578	26	29.01	25.93	-3.08	0.568
	Speech problems – All	61	0.467	60	22.77	21.86	-0.91	0.642
	Extraction or implants	34	-0.264	33	20.92	21.57	0.65	0.794
	'osteoradionecrosis'	27	0.925	26	25.10	22.22	-2.88	0.364
	Trouble with social eating – All	63	-0.358	62	37.57	38.89	1.32	0.721
	Extraction or implants	36	-1.439	35	32.87	38.19	5.32	0.159
	'osteoradionecrosis'	27	0.571	26	43.83	39.81	-4.02	0.573
	Trouble with social contact – All	62	-2.243	61	11.94	16.88	4.95	0.029
	Extraction or implants	36	-2.981	35	10.37	18.15	7.78	0.005
	'osteoradionecrosis'	26	-0.274	25	14.10	15.13	1.03	0.787
	Less sexuality – All	59	-0.710	58	36.16	38.42	2.26	0.481
	Extraction or implants	34	0.955	33	32.84	28.92	-3.92	0.347
	'osteoradionecrosis'	25	-2.317	24	49.67	51.33	1.66	0.029

Table 5 (Continued)

	EORTC QLQ-H&N35	N	t	DF	T ₁	T ₂	T ₁ -T ₂	p
Single Item Scales	Teeth – All	63	3.178	62	57.14	41.27	-15.87	0.002
	Extraction or implants	35	2.487	34	69.52	51.43	-18.09	0.018
	'osteoradionecrosis'	28	1.950	27	41.67	28.57	-13.10	0.062
	Opening mouth – All	62	1.196	61	55.38	50.54	-4.84	0.236
	Extraction or implants	34	-0.387	33	47.06	49.02	1.43	0.701
	'osteoradionecrosis'	28	2.091	27	65.48	52.38	-13.10	0.046
	Dry mouth – All	64	2.116	63	60.94	53.13	-7.81	0.038
	Extraction or implants	36	1.464	35	63.89	55.56	-8.33	0.152
	'osteoradionecrosis'	28	1.652	27	57.14	50.00	-7.14	0.110
	Sticky saliva – All	61	0.942	60	38.25	33.88	-4.37	0.350
	Extraction or implants	34	0.463	33	33.33	30.39	-2.94	0.646
	'osteoradionecrosis'	27	0.895	26	44.44	38.27	-6.17	0.379
	Coughing – All	62	0.388	61	29.03	27.42	-1.61	0.699
	Extraction or implants	35	1.244	34	39.05	31.43	-7.62	0.222
	'osteoradionecrosis'	27	-1.224	26	16.05	22.22	6.17	0.232
	Felt ill – All	63	1.183	62	25.40	21.16	-4.24	0.241
	Extraction or implants	36	1.313	35	29.63	23.15	-6.48	0.198
	'osteoradionecrosis'	27	0.238	26	19.75	18.52	-1.28	0.814
	Pain killers – All	63	0.903	62	74.60	69.84	-4.76	0.370
	Extraction or implants	36	-0.813	35	66.67	72.22	5.55	0.422
	'osteoradionecrosis'	27	2.431	26	85.19	66.67	-18.52	0.022
	Nutritional supplements – All	64	-0.830	63	31.25	35.94	4.69	0.410
	Extraction or implants	36	-0.813	35	25.00	30.56	5.56	0.422
	'osteoradionecrosis'	28	-0.372	27	39.29	42.86	3.57	0.713
	Feeding tube – All	64	1.000	63	14.06	10.94	-3.12	0.321
	Extraction or implants	36	1.435	35	11.11	5.56	-5.55	0.160
	'osteoradionecrosis'	28	0.000	27	17.86	17.86	0.00	1.000
	Weight loss – All	64	0.000	63	21.88	21.88	0.00	1.000
	Extraction or implants	36	-1.673	35	11.11	22.22	11.11	0.103
	'osteoradionecrosis'	28	1.441	27	35.71	21.43	-14.28	0.161
	Weight gain – All	63	-0.275	62	15.87	17.46	1.59	0.784
	Extraction or implants	35	0.000	34	20.00	20.00	0.00	1.000
	'osteoradionecrosis'	28	-0.441	27	10.71	14.29	3.58	0.663

N is the number of participants data used in the calculation, t is the critical value used in combination with degrees of freedom to assess significance, DF is degrees of freedom, which is the number of scores that are free to vary in calculating the statistic. p is the level of significance. T₁ = time point 1, T₂ = time point 2. For all items and scales, high scores indicate more problems.

attributed to the surgery experienced by the participants.

EORTC QLQ-C30 is a well developed, reliable general QoL instrument for cancer patients. Although this questionnaire is cancer specific it does have a global element, which shows significant improvement. The other two significant improvements (pain and dyspnoea) are explained more fully when judged by the sub-groups, with pain being significantly reduced in the extraction or implant

group while no significant differences were found in the 'osteoradionecrosis' group.

The head and neck cancer specific module EORTC QLQ-H&N35 was developed with the input of patients and has high content validity and sensibility. It has proved to be sensitive to change in this study indicating significant change in social contact, teeth, opening mouth, dry mouth, pain killers and sexuality. As previously noted, changes in this study could

be attributed in part to the surgery undergone by these patients. The improvement in relation to teeth may be connected to the removal of carious teeth and this is born out by the significance being evident in the extraction and implant sub-group. The positive change in 'opening mouth' in the debridement group may be due to a reduction in trismus. What brings about the change in trismus is an interesting question. It could be that the surgical intervention has facilitated greater mobility, allowing mouth opening with more comfort. Another explanation is that participants experience a loosening of fibrotic tissue following HBO₂. Further investigation with a similar patient group not having a surgical intervention, could help answer this question. The improvement in relation to teeth may be connected to the reduction in xerostomia and the removal of carious teeth. The authors could not identify any intervention, other than HBO₂, that would have significantly improved the patients with 'dry mouth'. This could mean that the patients had a greater amount or improved consistency

Table 6. Follow-up information from participants

	Dental extraction or implant placement	'osteoradionecrosis'
Discharge or lost to follow up	4	2
Healed	31	15
Ongoing problems	2	4
Occasionally shedding sequestra	1	0
Improved or healing slowly	0	2
Cancer recurrence or died	0	5
Total in each group	38	28

PB conceived the idea for the study. SAH, PB and DC contributed to the design and planning of the research. SAH was responsible for data collection. SAH analysed the data. SAH wrote the first draft of the manuscript. PB coordinated funding for the project. All authors edited and approved the final version of the manuscript.

of saliva, which allows for a greater protection of the teeth. This aspect has significant potential implication for the long-term health of patients' oral cavities and requires further investigation. The reduction in social contact and sexuality is not surprising given the requirement for most of the patients to be away from home during their treatments, up to three weeks at a time. This would be best addressed by the provision of more hyperbaric facilities, therefore allowing patients to return home each day.

In conclusion, this study suggests that a combination of HBO₂ and surgery contributes to an improved QoL in these patients and thus psychological status of the patient over the course of their continued care.

Acknowledgements. Our thanks to the British Hyperbaric Society for their funding which allowed us to purchase questionnaires at the start of the research.

References

- ANNANE D, DEPOND J, AUBERT P, VILART M, GEHANNON P, GAJDOS P, CHEVRET S. Hyperbaric oxygen therapy for radionecrosis of the jaw: a randomized, placebo-controlled, double-blind trial from the ORN96 study group. *J Clin Oncol* 2004; 22: 4893-4900.
- BJORDAL K, AHLNER-ELMQVIST M, TOLLESON E, JENSEN AB, RAZAVI D, MAHER EJ, KAASA S. Development of a European Organization for Research and Treatment of Cancer (EORTC) questionnaire module to be used in quality of life assessments in head and neck cancer patients. EORTC Quality of Life Study Group. *Acta Oncol* 1994; 33: 879-885.
- BJORDAL K, BOYSEN M, JANNERT M. A Prospective Study of Quality of Life in Head & Neck Cancer Patients. Part II: Longitudinal Data. *Laryngoscope* 2001; 111: 1440-1452.
- CHANDU A, SMITH ACH, ROGERS SN. Health-Related Quality of Life in Oral Cancer: A Review. *J Oral Maxillofac Surg* 2006; 64: 495-502.
- DE GRAEFF A, DE LEEUW Jr, ROS WJ, HOEDJIK G, BLUHAM GH, WINNIBIST JA. Long-Term Quality of Life of Patients with Head & Neck Cancer. *Laryngoscope* 2000; 110: 98-106.
- DELANIAN S, PORCHER R, RUDANT J, LEFAIX J. Kinetics of Response to Long-Term Treatment Combining Pentoxifylline and Tocopherol in Patients with Superficial Radiation-Induced Fibrosis. *J Clin Oncol* 2005; 23: 8570-8579.
- D'SOUZA J, GORU J, GORU S, BROWN J, VAUGHAN ED, ROGERS SN. The influence of hyperbaric oxygen on the outcome of patients treated for osteoradionecrosis: 8 year study. *Int J Oral Maxillofac Surg* 2007; 36: 783-787.
- FAYERS P, AARONSON NK, BJORDAL K, GROENVOLD M, CURRAN D, BOTTOMLEY A, on behalf of the EORTC Quality of Life Group, EORTC QLQ-C30 Scoring Manual (3rd Edition). ISBN 2-930064-22-6. Brussels: EORTC; 2001: 1-86.
- FELDMER JJ. Hyperbaric oxygen for delayed radiation injuries. *Undersea Hyperb Med* 2004; 31: 133-145.
- FELDMER JJ, HAMPTON NB. A systematic review of the literature reporting the application of hyperbaric oxygen prevention and treatment of delayed radiation injuries: an evidence based approach. *Undersea Hyperb Med* 2002; 29: 4-30.
- GRANSTRÖM G. Placement of Dental Implants in Irradiated Bone: The Case for Using Hyperbaric Oxygen. *J Oral Maxillofac Surg* 2006; 64: 812-818.
- HANCOCK PJ, ERSTEIN JB, SADLER GR. Oral and dental management related to radiation therapy for head and neck cancer. *J Can Dent Assoc* 2003; 69: 585-590.
- HODDER SC, EVANS RM, PATTON DW, SILVESTER KC. Ultrasound and fine needle aspiration cytology in the staging of neck lymph nodes in oral squamous cell carcinoma. *Br J Oral Maxillofac Surg* 2000; 38: 430-436.
- KELLER EE. Placement of Dental Implants in the Irradiated Mandible: A Protocol Without Adjunctive Hyperbaric Oxygen. *J Oral Maxillofac Surg* 1997; 55: 972-980.
- LARSEN PE. Placement of Dental Implants in the Irradiated Mandible: A Protocol Involving Adjunctive Hyperbaric Oxygen. *J Oral Maxillofac Surg* 1997; 55: 967-971.
- LENT SOMA tables. *Radiother Oncol* 1995; 35: 17-60.
- MARX RE, AMES JR. The use of hyperbaric oxygen therapy in bony reconstruction of the irradiated and tissue-deficient patient. *J Oral Maxillofac Surg* 1982; 40: 412-420.
- MARX RE, JOHNSON RP, KLINE SN. Prevention of 'osteoradionecrosis': a randomized prospective clinical trial of hyperbaric oxygen versus penicillin. *J Am Dent Assoc* 1985; 111: 49-54.
- MYERS RA, MARX RE. Use of hyperbaric oxygen in post radiation head and neck surgery. *NCI Monogr* 1990; 9: 151-157.
- MYKLETUN A, STORDAL E, DAHL AA. Hospital Anxiety and Depression (HAD) scale: factor structure, item analyses and internal consistency in a large population. *Br J Psychiatry* 2001; 179: 540-544.
- OFFICE OF NATIONAL STATISTICS, Cancer Statistics registrations: Registrations of cancer diagnosed in 2005, England. London: Office of National Statistics 2008.
- O'SULLIVAN B, GULLANE P, IRISH J, NELIGAN P, GENTILI F, MAHONEY J, SELLMANN S, CATTON C, WALDRON J, BROWN D, WITTERICK I, FREEMAN J, BELL R. Preoperative radiotherapy for adult head and neck soft tissue sarcoma: assessment of wound complication rates and cancer outcome in a prospective series. *World J Surg* 2003; 27: 875-883.
- ROGERS SN, LOWE D, FISHER SE, BROWN JS, VAUGHAN ED. Health-related quality of life and clinical function after primary surgery for oral cancer. *Br J Oral Maxillofac Surg* 2002; 40: 11-18.
- ROGERS SN, GWANNE S, LOWE D, HUMPHRIS G, YUEH B, WEYMULLER EA. The addition of mood and anxiety domains to the University of Washington Quality of life scale. *Head Neck* 2002; 24: 521-529.
- SULAIMAN F, HURYN JM, ZLOTOW LM. Dental extractions in the irradiated Head and Neck Patient: A retrospective analysis of Memorial Sloan-Kettering Cancer Center protocols, criteria, and end results. *J Oral Maxillofac Surg* 2003; 61: 1123-1131.
- TROTTI A, JOHNSON DJ, GWEDD C, CASEY L, SAUDER B, CANTOR A, PEARLMAN J. Development of a head and neck comparison module for the quality of life-radiation therapy instrument (QOL-RTI). *Int J Radiat Oncol Biol Phys* 1998; 42: 257-261.
- WAKE JE Jr. Standards for validating health measures: definition and content. *J Chronic Dis* 1987; 40(6):473-480.
- WAKE JE Jr., SNOW KK, KOSINSKI M, GANDEK B. SF-36 Health Survey. Manual and Interpretation Guide. 2nd ed. Lincoln Rhode Island QualityMetric Incorporated, 2002.
- ZELEFSKY MJ, GAYNOR J, KRAUS D, STRONG EW, SHAH JP, HARRISON LB. Long-term subjective functional outcome of surgery plus postoperative radiotherapy for advanced stage oral cavity and oropharyngeal carcinoma. *Am J Surg* 1996; 171: 258-261.
- ZIGMOND AS, SNAITH RP. The Hospital Anxiety and Depression Scale. *Acta Psych Scand* 1984; 67: 361-370.

Address:
Sam A Harding
Hyperbaric Medical Centre
Derriford
Plymouth
Devon
PL6 8BU
UK
Tel.: +44 0 1752 209999
Fax: +44 0 1752 209115
E-mail: sharding.jh@googlemail.com

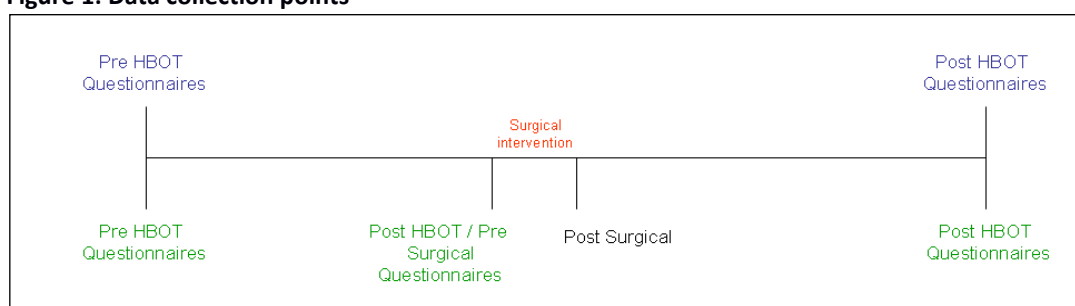
4.2.4 Study Limitations

4.2.4.1 Data collection time points

Over the duration of the work reported in paper 4, it became evident that I had not optimised the data collection time points. Figure 1 shows, in the blue, when I collected the data reported. What I found after approximately a year was that on occasion people completed their pre-surgical course of HBOT, and then because of the level of healing or general improvement in their condition, did not have surgery. One of two things would then happen; they returned for more HBOT to consolidate their healing, or did not return, as their consultant felt additional treatments were not necessary. In the latter case I did not have the opportunity to collect 'post HBOT' measures.

To address this, I obtained an amendment to the study protocol and undertook data collection at the three time points in green in figure 1. It was decided not to ask people to complete a measure post-surgery because this can be as little as one day and usually no more than a week from when they completed their pre-op HBOT.

Figure 1: Data collection points



This data has yet to be written up for publication, but it is anticipated that for at least some variables, there would be an improvement between pre-HBOT and Post-HBO/Pre-surgery and then a further improvement at the end of the subsequent series of HBOT. It would be difficult to unpick what improvements are attributable to the HBOT and what to the surgery. A control group would aid our understanding of those relationships.

4.2.4.2 Control groups

As DDRC is a charity and outside the NHS, it is impossible to get a control group in the type of pragmatic recruitment study undertaken in papers 4 and 5. I could not choose to not treat people referred to the service to see how they did and how their QoL changed, by receiving standard non-HBOT treatment or natural progression of

their disorder. I did not have access to the NHS databases to find matched participants, and with the referral process, we were unable to undertake an extended baseline assessment to establish the stability of the respondents QoL prior to their HBOT.

There are ways to add a HBOT control in for the treatment, in a similar way to giving a Placebo. This would be to undertake research with sham treatments. The goal of a sham treatment is to ensure that patients and investigators are unable to distinguish sham from actual HBOT (thereby removing a potential placebo effect), while the sham procedure must not have any effect on the disease being treated. Because patients have to clear (or pop) their ears when pressure is increased in HBOT, sham therapy also has to use pressure to create/mimic this experience. However, increasing the atmospheric pressure has an effect on the partial pressures of gases, potentially causing the sham treatment to become an active agent. This dilemma, together with considerations regarding practicality, safety and blinding, has resulted in different strategies being used in various trials over the years, each with their own advantages and disadvantages.

- 1) use of a lower pressure than that of the hyperbaric oxygen group, while breathing 21% oxygen
- 2) use of the same pressure as the hyperbaric oxygen group, while breathing an adjusted percentage of oxygen
- 3) use of the same pressure as the hyperbaric oxygen group, while breathing 21% oxygen

Once again because the data was being collected on patients referred for treatment, rather than recruited into a research trial, it was not possible to use any of these control groups either. Although, understanding how these controls would work and their implications is vital if undertaking a distinct research project where a control would be included and costed into the grant.

4.2.5 Contribution to knowledge and autoethnographic issues

4.2.5.1 Contribution to knowledge

At the time we started data collection (2001), only one study of HBOT and HNC had included any measure of QoL (Chougule et al, 1999) and by the time this paper was published (2008) only two more had been published (Gelach et al 2008; Schoen et al

2007), both with significantly smaller sample sizes (21 and 26 respectively).

Therefore, this paper gave greater data to allow for power calculations to be undertaken in future research. It also provides some idea of the patient-identified added bonus provided by HBOT, as they reported improvements in factors such as xerostomia, social contact and shoulder movement, none of which were areas directly related to the purpose of their original referrals.

Appendix 9 has an overview of how the research undertaken in paper 4 was disseminated prior to the paper's publication. It also provides a list of papers where paper 4 has been cited, and this is followed by a quality appraisal of the paper.

4.2.5.2 Autoethnographic issues

This was the first research involving 'patients' that I undertook. I designed, gained ethical approval, ran and reported it from start to finish. What this clearly showed me was that clinical research can be embedded into everyday practice. It need not be a distinctly separate piece of work with specific people only allowed or enabled to undertake tasks. There are specialist skills and knowledge involved but there were also many opportunities for research to be a part of 'normal' everyday clinical practice, just as with every other specialist role within the NHS.

I feel that embedding research is a proactive way to ensure the findings are almost immediately ready to be used in practice and that these findings have emerged directly from day-to-day clinical practice. In discussion with some people employed as researchers this view has been strongly opposed, with them voicing the opinion that research cannot be done without specific funding and personnel. I am not sure to what extent their views are to do with protecting their thoughts and beliefs of role identity as allied health professional researchers, or if they believe that research cannot be done on a day-to-day basis without specific funding. I, however, continue to believe that research is a team activity that can be incorporated into everyday clinical practice; although I am positive this discussion will be had for many years to come.

While undertaking this research, I was also working clinically for the first time. The people we treated while I was at DDRC helped me understand what it is like to have HNC, their extended journey through diagnosis to treatment to the long term side effects of treatment and the impact this has on them biologically, psychologically and socially. They also told me how, me asking them permission to help them when they

appeared they needed assistance, made them feel empowered, and how giving them back a sense of self whilst in a medical environment was something they had not realised they had lost. These experiences and many more have made me want to ensure that people/patients I work with must be asked about the projects to ensure they are relevant to them, and what problems I might be running into from their perspective, and what significant issues clinicians are overlooking. As a minimum I am informed by the national standards outlined by NIHR Involve (<https://www.invo.org.uk/posttypepublication/national-standards-for-public-involvement/>), but more often than not the people I am working with have their own distinct ideas about how to move forward and the processes they want put in place.

4.2.6 Future research questions

As mentioned, the involvement of patients and the public was driven home during this work. It is important to ensure that the research is relevant and seen as important by those people who will potentially be participants. It also has the beneficial effect of making it easier 'to sell' projects to potential participants, thereby maximising recruitment. I have therefore actively sought out support groups or willing individuals to inform the development and running of research that I have developed.

Regarding the research reported in paper 4, this led to a modification of the data collection protocol and data was collected post HBOT and pre surgery. As mentioned above, some patients did not receive a surgical intervention during their HBOT. This meant that their data needed to be considered differently and directly influenced the production of paper 5, with a strictly defined sub-group of people.

4.3 Paper 5 - Clinical experience leading to the research work and production of the paper

Whilst undertaking the research presented in paper 4, it became apparent that some patients were either referred for treatment of Type III Osteoradionecrosis (ORN) or spontaneous ORN. This type of ORN can occur any time after radiotherapy, but usually has an onset between 6 months and 2 years, and occurs without any obvious surgical or traumatic event. This differs from the people represented in paper 4, as they were being treated prophylactically, in order to minimise the onset or progression of ORN due to having dental extractions or implants in an irradiated area.

It became clear that this group of people, with Type III ORN, had a different experience than the other people who had had HNC, prior to attending DDRC. They tended to have some side effects of treatment such as xerostomia or trismus which they were managed symptomatically with e.g. saliva substitutes, or through behavioural changes, such as soft food diets and cutting food into smaller pieces. At some point they had experienced sudden pain or discomfort in their jaw, which subsequently transpired to be a fracture in their mandible or less frequently their maxilla. Their consultants often were reluctant to intervene surgically due to the risk of exacerbating their problem, as surgical plating requires 'good quality' bone to anchor the plate to, and people with ORN in their jaw do not have this. Therefore, surgery can lead to greater side effects, and potentially in some of the worst cases, this can lead to patients having to have their mandible fully removed (mandibulectomy) and being left with what is known as the 'Andy Gump' deformity (Figure 2).

This was a very different trajectory to the participants represented in paper 4. Although their condition had the potential to progress negatively, this was thought to be over tens of years. However, those with Type III ORN, had experienced a sudden change with a potentially very quick progression of their condition leading to major surgery and significant life changing events. From these conversations and their difference in treatment patterns it was clear that a separate paper/research was needed.

Figure 2: Severe deformity



4.3.1 Research Questions

What changes in QoL, reported via a battery of questionnaires, are experienced by people receiving HBOT for Type III ORN following treatment for HNC?

4.3.2 Study Design

The same study design was used for paper 5 and paper 4, reported in section 4.2.2.

4.3.3 Effects of Hyperbaric Oxygen Therapy on Quality of Life in Maxillofacial Patients with Type III Osteoradionecrosis

Effects of Hyperbaric Oxygen Therapy on Quality of Life in Maxillofacial Patients With Type III Osteoradionecrosis

Sam Harding, MSc, MPbil,* David Courtney, BDS, BM,†
Simon Hodder, MBBS, BDS,‡ and Philip Bryson, MBBS§

Purpose: Over a 4-year period, 18 patients with type III osteoradionecrosis that developed an average of 55 months after radiotherapy treatment for head and neck cancers were referred for hyperbaric oxygen therapy (HBO₂).

Materials and Methods: Participants completed a questionnaire battery before and after HBO₂, including the European Organization for Research and Treatment of Cancer (EORTC) Core 30, the EORTC Head and Neck 35, and the Medical Outcomes Short Form 36.

Results: The EORTC Core 30 questionnaire indicated significant improvements in "emotional functioning" and "insomnia" ($P \leq .01$ and $P \leq .01$). An improvement also was found in the "social eating" ($P \leq .01$) and "teeth" ($P \leq .01$) domains of the EORTC Head and Neck 35 questionnaire. These beneficial outcomes might be explained in part by the social environment of being in a specific treatment group with similar patients. However, the Medical Outcomes Short Form 36 indicated a significant decrease in "social functioning" ($P \leq .01$). The patient group in this study did not undergo any surgical intervention between the 2 time points and no other interventions could be connected with the improvements, particularly in relation to "teeth." In addition, clinical follow-up confirmed the stabilization of the patients' clinical conditions.

Conclusions: The findings of this study support the hypothesis that HBO₂ has positive physiologic and psychological effects on some factors for this patient group.

© 2012 American Association of Oral and Maxillofacial Surgeons. Published by Elsevier Inc. All rights reserved.

J Oral Maxillofac Surg 70:2786-2792, 2012

Maxillofacial carcinomas are the eighth most common form of cancer in the UK population.¹ Surgery, radiotherapy, and chemotherapy alone or in combination are the main treatment modalities. Despite the life-saving abilities of these treatments, there are some serious side effects. These include mucositis, fibro-

sis, xerostomia, trismus, and, in approximately 2% of cases, osteoradionecrosis (ORN) and soft tissue radionecrosis.² These side effects are, for the most part, deemed irreversible and have a significant demonstrable negative effect on a patient's quality of life (QoL).³ In some cases, surgery is considered part of the long-term treatment of the patient, but surgical intervention in a heavily irradiated field may result in delayed wound healing, dehiscence, or infection.^{4,5} These factors combined with a patient's other comorbidities affect consultants' decisions on ongoing treatment.

Hyperbaric oxygen therapy (HBO₂) is increasingly accepted as a treatment for radiation proctitis,⁶ and it has been suggested as a medical treatment for soft tissue radionecrosis in other parts of the body.⁷⁻⁹ However, HBO₂ is not generally used as a stand-alone treatment for ORN because dead bone needs to be removed surgically.

ORN develops in 3 well-established clinical scenarios and produces 3 types: type I occurs when teeth are removed from a jaw to be radiated and fewer than 21 days are allowed for tissue recovery and healing

*Psychologist, DDRC, The Hyperbaric Medical Centre, Derriford, Plymouth, Devon, UK.

†Consultant Oral and Maxillofacial Surgeon, Maxillofacial Department, Derriford Hospital, Derriford, Plymouth, Devon, UK.

‡Consultant Oral and Maxillofacial Surgeon, Department of Oral and Maxillofacial Surgery, Morriston Hospital, Morriston, Swansea, UK.

§Medical Director of Diving Services, Abermed Ltd, Aberdeen, UK.

The British Hyperbaric Association funded the purchase of the quality-of-life questionnaires.

Address correspondence and reprint requests to Ms Harding: DDRC, The Hyperbaric Medical Centre, Derriford, Plymouth, Devon, PL6 8BU, UK; e-mail: sharding.jb@gmail.com

© 2012 American Association of Oral and Maxillofacial Surgeons. Published by Elsevier Inc. All rights reserved.

0278-2391/12/7012-0278\$36.00/0

<http://dx.doi.org/10.1016/j.joms.2012.04.011>

before commencing radiotherapy; type II occurs years after radiotherapy and is a result of external or surgical trauma; and type III occurs spontaneously after radiotherapy and is not related to any trauma.¹⁰ In maxillofacial patients with types I and II ORN, HBO₂ is used in a regime that sandwiches surgery according to the Marx protocols,¹¹⁻¹⁴ and this treatment modality has been shown to have a positive effect on QoL.^{15,16} In the present research, the authors were interested in type III ORN, ie, that which occurs spontaneously. Whatever the presentation, surgeons are generally keen to avoid or minimize surgery, if possible, because of the potential to exacerbate the problem and the patients' comorbidities.

In this report, the authors describe the changes in QoL reported in a questionnaire battery by patients undergoing HBO₂ as a treatment for type III ORN.

Materials and Methods

ETHICS

Ethical approval was granted from the local research ethics committee according to British Psychological Society guidelines and the Declaration of Helsinki. The study was explained to potential recruits from an information sheet and questions were answered. Written consent was obtained.

PARTICIPANTS

Eighteen patients (13 men; mean age, 63.6 yr) referred for HBO₂ after radiotherapy for head and neck cancer (HNC) were recruited to complete a questionnaire battery before and after HBO₂. The questionnaires before HBO₂ were completed after a medical assessment for fitness to undertake HBO₂ and before a patient's first treatment on the same day. The questionnaires after HBO₂ were undertaken after the last HBO₂ before formal discharge from the hyperbaric unit. There was an average of 28 days between these 2 time points.

The average body mass index for the participants was 24.17 kg/m² (standard deviation, 4.01 kg/m²), which is within the "normal" range. Demographic data were collected from the patients' hyperbaric medical notes and are presented in Table 1. Table 1 also includes the referring consultants' review of the patients' status 2 years after HBO₂ had been completed.

INCLUSION AND EXCLUSION CRITERIA

All patients were older than 18 years and had English as their first language. None of the patients had previously undergone HBO₂. Patients were referred with type III ORN that had been confirmed by orthopantomogram and clinical examination. Referral was made to minimize the need for surgical intervention.

HBO₂ REGIME

Patients received 29 to 49 therapies (mean, 34.0; standard deviation, 6.1) in a multiplace chamber at the Hyperbaric Medical Centre (Plymouth, UK). All participants underwent HBO₂ twice a day at 2.2 Atmospheres Absolute (12 m) for 45 minutes, an air break for 5 minutes, and then another 45 minutes (in total, 90 min breathing 100% oxygen) for 5 days a week (Fig 1). The daily treatments were separated by a minimum of 3.5 hours. Oxygen was delivered through an Amron Oxygen Treatment Hood (Vista, California) or a Sea-Long Series 7000 Mask (Louisville, Kentucky).

THE QUESTIONNAIRE BATTERY

Currently, there are no QoL questionnaires designed specifically for use in hyperbaric medicine. The measurements used in this research were developed and validated in settings such as outpatient clinics and in the hospital environment and therefore were deemed valid and appropriate for the assessment of change in this study. Two questionnaires were used: the Medical Outcomes Short Form 36 (SF-36)¹⁷ and the European Organization for Research and Treatment of Cancer (EORTC)¹⁸ Core 30 (QLQ-C30) with its subscale, the EORTC Head and Neck 35 (QLQ-HN35).¹⁹ These questionnaires were outlined in a previous article that assessed patients with HNC undergoing HBO₂ perioperatively.¹⁶

The participants completed the questionnaires independently and unsupervised.

ANALYSIS METHOD

The primary statistical method used in this research was the Wilcoxon sign-rank test because of the small sample. To account for the number of subscales within the measurements used, statistical significance was set to $P \leq .01$.

Results

Participants had completed their cancer treatment on average 55 months before starting their HBO₂.

Using the SF-36, improvements (although not to a significant level) were found across all domains except for "social functioning," which showed a significant decrease (Table 2).

Significant changes were evident using the EORTC QLQ-C30 (Table 3) in "emotional functioning" and "insomnia." As with the SF-36, most subscales showed improvement trends but did not reach significance at $P \leq .01$.

This pattern of improvement continued in the domains of the EORTC QLQ-HN35, where significant amelioration in the domains of "social eating" and "teeth" were found (Table 4).

Table 1. DEMOGRAPHIC DATA

Patient Number	Gender	T	N	M	Number of HBO ₂ Treatments	Absorbed Radiation (Gy)	Time Since Radiotherapy (mo)	Smoking (Cigarettes/day)	Alcohol (U/wk)	BMI on Admission	Referring Clinicians' Outcome Reports 2 yr After HBO ₂
1	Female	—	—	—	30	76	72	0	0	25	No requirement for surgical intervention
2	Male	2	0	ND	36	55	23	10	0 (reformed alcoholic, quit 5 yr previously)	24	Did very well but within 6 months needed surgery
3	Male	4	0	ND	36	60	18	>20	10-20	21	ORN stabilized, no surgery needed
4	Male	1	0	0	36	60	12	>20	20-40	27	ORN showed good response, stable
5	Male	4	1	ND	30	54	48	Quit 4-5 yr previously	1	22	ORN stable, no surgery planned
6	Female	4	2	ND	35	60	30	Quit 1-5 yr previously	—	—	ORN did very well
7	Female	—	—	—	38	—	192	20 before quitting 16 yr previously	Occasional	21	ORN stabilized, no surgery planned
8	Male	2	0	ND	38	—	120	0	14	25	ORN stable but needed minor debridement
9	Male	2	1	ND	29	—	12	15-20	—	22	ORN stable but needed surgery and HBO ₂ 2 yr later
10	Male	1	0	ND	40	60	56	Quit pipe smoking 4 yr previously	18	26	ORN stabilized, no surgery required
11	Male	2	0	ND	29	—	60	4	Occasional	—	ORN showed good outcome but remains with problems
12	Male	4	2	ND	30	—	49	10	3	20	ORN stabilized, no need for surgery
13	Male	1	1	ND	29	—	96	Quit 22 yr previously	0	35	Minor debridement required
14	Female	—	—	—	29	42	84	—	—	—	ORN appears to be stable
15	Male	2	0	ND	30	—	60	2	3	19	ORN did well but developed recurrent cancer 18 mo later
16	Male	1	0	0	46	90	36	Quit 20 yr previously	Occasional	29	ORN did well but required surgery some 18 mo later
17	Male	4	0	ND	29	—	18	0	1	23	ORN stabilized, no need for surgery
18	Female	4	0	ND	49	50	7	0	Very occasional	26	ORN stabilized, no need for surgery

Abbreviations: —, data not provided by referring clinician; BMI, body mass index; HBO₂, hyperbaric oxygen therapy; M, metastatic stage; N, nodal stage; ND, not determined; ORN, osteoradionecrosis; T, tumor stage.

Harding et al. Conservative HBO₂, Type III ORN, and QoL. *J Oral Maxillofac Surg* 2012.

Discussion

QoL measurements have been used widely in the assessment of patients with HNC malignancy. They are a valuable tool because these cancers and the treatment that patients receive can have a significant impact on individuals' QoL.

Given the number of variables previously shown to have an impact on health-related QoL in this patient group and the variation between patients and missing data,³ it is not surprising that so few factors reached significance in this study. However, the trend throughout the data of an improvement does suggest a beneficial effect of HBO₂ on QoL.

As with the patients in this study (Table 1), it is sometimes reported to Diving Diseases Research Centre (DDRC) that patients referred for HBO₂ have not undergone surgery owing to a significant improvement in their condition. This explains the spread of the number of treatments in those having 29 to 30 being referred for pre- and postoperative HBO₂ and not returning to DDRC for postoperative treatment

because the referring consultants judged that surgery was unnecessary. Those participants having more than 30 therapies were those returning for what would have been postoperative treatments, but were actually therapies to consolidate healing without surgery. The "preoperative" series of HBO₂ had improved the patients' condition to the extent that the referring consultants did not want to operate but judged some further HBO was needed.

Because of the nature and workload of the hyperbaric unit where these data were collected (charity outside the National Health Service, UK), it is often difficult to obtain all the patient data that are requested, such as radiotherapy dose (Table 1). This is due to the patient files being incomplete across organizations, oncology units using separate computer systems from the other referring hospitals, and the expense (financially and in time) of patient note reviews. In addition to these issues, it was impossible to include control groups because of funding issues. With this in mind, a repeated measures design was

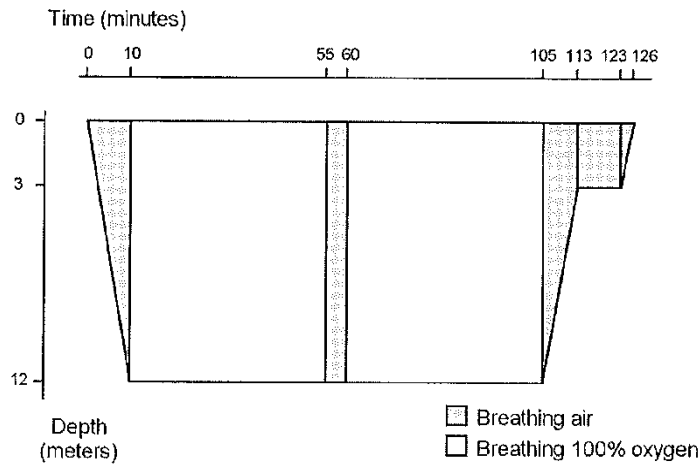


FIGURE 1. Treatment profile.

Harding et al. Conservative HBO₂ Type III ORN, and QoL. *J Oral Maxillofac Surg* 2012.

chosen for the study, providing some degree of internal control to the data collection. A randomized placebo-controlled trial is, of course, the gold standard methodology and minimal air compression is an effective blinding tool for patients enrolled in hyperbaric trials.²⁰ Multicenter studies with this patient cohort looking at the effect of HBO₂ and surgery are underway using this methodology. However, there is considerable operational expense, and with the addi-

tion of ethical and logistical considerations, this can more than double the costs.

Previous work by the present authors in patients with HNC undergoing HBO₂ perioperatively attributed improvements in QoL to the combination of HBO₂ and surgery.¹⁶ That work was criticized for the risk of possible type I errors owing to the large amount of data being presented.²¹ Some of the same scales were used in the present work and, therefore, to a limited extent the same criticism could be made. A Bonferroni correction could be used to correct for this. However, the Bonferroni correction is a very conservative measurement and would lead to an increased risk of type II errors (rejecting significances that are actually present).²² Therefore, a more conservative level of significance ($P \leq .01$) was chosen. Power calculations performed on the data collected suggest that a sample size of 50 patients completing questionnaires before and after HBO₂ would allow a greater understanding of the effect of treatment and patient variables, including age and gender. Larger numbers would be required to investigate the influence of factors such as smoking/alcohol status and body mass index. These latter factors may be of interest because they have previously been shown to influence the onset of ORN.²³⁻²⁵

Many facets of QoL approach normal levels after the initial decreases around the time of treatment.³ In the present research, the mechanism of the referral and treatment process prevented the authors from assessing the longitudinal stability of QoL measurements in this patient group; however, the mean average assessment of the patients was 55 months (4 yr 7

Table 2. MEDICAL OUTCOMES SHORT FORM 36 SCORES

	n	T1, Mean (SD)	T2, Mean (SD)	T1 vs T2
Physical functioning	18	39.9 (11.1)	40.2 (12.0)	0.3
Role—physical	18	28.4 (10.0)	32.0 (11.8)	3.6
Bodily pain	18	46.2 (10.4)	47.8 (10.7)	1.6
General health	18	39.3 (12.5)	44.7 (9.6)	5.4
Vitality	18	46.0 (10.4)	47.9 (11.1)	1.9
Social functioning	18	62.0 (24.8)	42.8 (12.4)*	-19.2
Role—emotional	18	23.5 (14.2)	32.5 (16.8)	9.0
Mental health	18	46.7 (12.0)	51.0 (10.5)	4.3

NOTE. A higher score indicates a better quality of life; therefore, a negative difference indicates a decline in quality of life.

Abbreviations: n, number of participants' data used in the calculation; SD, standard deviation; T1, before hyperbaric oxygen therapy; T2, after hyperbaric oxygen therapy.

*Significant at .01.

Harding et al. Conservative HBO₂ Type III ORN, and QoL. *J Oral Maxillofac Surg* 2012.

Table 3. EUROPEAN ORGANIZATION FOR RESEARCH AND TREATMENT OF CANCER CORE 30 QUESTIONNAIRE DATA

	n	T1, Mean (SD)	T2, Mean (SD)	T1 vs T2
Global health status/QoL*	18	57.9 (14.7)	69.6 (12.5)	11.7
Physical functioning*	18	72.2 (17.9)	75.7 (19.0)	3.5
Role functioning*	18	70.4 (29.5)	64.3 (32.0)	-6.1
Emotional functioning*	18	63.4 (21.2)	81.0 (18.6) [‡]	17.6
Cognitive functioning*	18	61.1 (22.9)	75.6 (22.2)	14.5
Fatigue [†]	18	40.1 (21.9)	41.8 (23.2)	1.7
Nausea and vomiting [†]	18	8.3 (11.8)	3.6 (7.1)	-4.7
Pain [†]	18	25.9 (24.4)	25.0 (23.3)	0.9
Dyspnea [†]	18	33.3 (25.6)	23.8 (30.5)	-9.5
Insomnia [†]	18	50.0 (34.8)	31.0 (33.2) [‡]	-19.0
Appetite loss [†]	18	29.6 (36.0)	15.4 (32.2)	-14.2
Constipation [†]	18	18.5 (28.6)	14.3 (25.2)	-4.2
Diarrhea [†]	18	0.0 (0.0)	2.4 (8.9)	2.4
Financial impact [†]	18	27.8 (30.8)	14.3 (21.5)	-13.5

Abbreviations: n, number of participants' data used in the calculation; QoL, quality of life; SD, standard deviation; T1, before hyperbaric oxygen therapy; T2, after hyperbaric oxygen therapy.

*Higher score indicates better function.

†Higher score indicates more symptoms.

‡Significant at .01.

Harding et al. *Conservative HBO₂, Type III ORN, and QoL*. *J Oral Maxillofac Surg* 2012.

mo) after treatment. Chandu et al³ suggested that short-term morbidity can be generally stable after 1 year, and nearing precancer levels by 3 years. It is reasonable to assume the QoL in these patients to be stable before HBO₂ and, hence, that the changes found in this study are due to HBO₂ and the experiences they had while at the hyperbaric medical center undergoing treatment.

The SF-36 is often used in medical trials as a stand-alone measurement and has been shown to be reliable and valid in a clinical setting.^{17,26} In a previous study on HBO₂ and a similar patient group, the SF-36 failed to identify any changes in QoL.¹⁶ With the present patient group, the SF-36 indicated a decrease in reported "social functioning." This most likely was because the patients spent an extended period away from home and, therefore, their family and social activities. The same explanation can be given about the decrease in sexuality identified in the EORTC QLQ-HN35 (although not significant). The other SF-36 domains identified positive trends (Tables 2, 4), suggesting that there is a significant positive effect on the patients being in a group of people who have experienced a similar illness, treatments, and side effects.

Anecdotal reports from patients at the Hyperbaric Medical Centre in Plymouth suggest that improvements in mouth opening and the physical sensations relating to this change make talking more comfortable. A qualitative study may provide a greater insight into the specifics of the positive effects, but the authors postulate that this improved ability to talk to and see others coping with similar issues gives patients the resources to be more emotionally and mentally able. There are limitations to this hypothesis. The Hyperbaric Medical Centre does not treat only patients with ORN after treatment for HNC; they also treat conditions such as diabetic foot ulcers. This variation in patient cohort means that there were members of the study group who received HBO₂ but did not have other patients with HNC with whom to talk and socialize. It was also the case that some patients with HNC did not engage with their patient peers. This means that the delivery of the HBO₂ was carefully controlled and can be assessed and evaluated, but socialization and its mechanism of action is more complex and the number of participants in this

Table 4. EUROPEAN ORGANIZATION FOR RESEARCH AND TREATMENT OF CANCER HEAD AND NECK 35 QUESTIONNAIRE DATA

	n	T1, Mean (SD)	T2, Mean (SD)	T1 vs T2
Pain	18	35.3 (25.4)	32.1 (20.9)	-3.2
Swallowing	18	27.9 (20.6)	19.4 (17.9)	-8.5
Senses problem	18	40.2 (36.8)	35.9 (41.9)	-4.3
Speech problems	18	28.8 (19.7)	22.2 (21.8)	-6.6
Trouble with social eating	18	52.3 (23.5)	30.8 (26.4) [*]	-21.5
Trouble with social contact	18	24.7 (22.8)	22.6 (26.2)	-2.1
Less sexuality	18	52.0 (41.6)	65.3 (33.7)	13.3
Teeth	18	48.9 (43.4)	22.2 (30.0) [*]	-26.7
Opening mouth	18	74.5 (32.3)	57.1 (35.6)	-17.4
Dry mouth	18	58.8 (41.7)	51.3 (44.3)	-7.5
Sticky saliva	18	49.0 (41.0)	56.6 (34.4)	7.6
Coughing	18	31.4 (18.5)	25.6 (30.9)	-5.8
Felt ill	18	19.6 (20.6)	15.4 (22.0)	-4.2
Pain killers	18	55.6 (51.1)	57.1 (51.4)	1.5
Nutritional supplements	18	44.4 (51.1)	28.6 (46.9)	-15.8
Feeding tube	18	5.6 (23.6)	0.0 (0.0)	-5.6
Weight loss	18	16.7 (38.3)	0.0 (0.0)	-16.7
Weight gain	18	11.1 (32.3)	28.6 (46.9)	17.5

NOTE: For all items and scales, high scores indicate more problems; therefore, a negative difference indicates an improvement in quality of life.

Abbreviations: n, number of participants' data used in the calculation; SD, standard deviation; T1, before hyperbaric oxygen therapy; T2, after hyperbaric oxygen therapy.

*Significant at .01.

Harding et al. *Conservative HBO₂, Type III ORN, and QoL*. *J Oral Maxillofac Surg* 2012.

study is insufficient to be able to make any generalizable conclusions about its impact on QoL.

The EORTC QLQ-C30 is a well-developed, reliable, general QoL instrument for patients with cancer. Although this questionnaire has cancer specific subscales, in the present case, the QLQ-HN35 and the global element (QLQ-C30) showed significant improvement in the ORN group. The decrease in "insomnia" may be explained by the improvement in "emotional functioning" (Table 3). Because insomnia is common in this patient group with psychiatric morbidity, the authors considered there may well be a link between these improvements.²⁷ This finding suggests that, even after completion of treatment, patients with cancer can benefit from group interaction and support.

The EORTC QLQ-HN35, like the other scales, produced data showing a positive trend for QoL across most domains. However, only 2 significant differences were "social eating" and "teeth" ($P \leq .01$ for the 2 comparisons; Table 4). The improvement in "social eating" may be explained by the informal patient interaction that occurs at the hyperbaric unit. Patients can talk about their condition and the problems that are affecting them, often leading to an exchange of problem solving, which includes attitudes toward eating in public and an increase in self-confidence. The change in relation to "teeth" cannot be explained by psychological factors. The domain within the EORTC QLQ-HN35 is a single item: Have you had problems with your teeth? After HBO₂ the patients reported significantly fewer problems than before treatment. No surgical or dental interventions had taken place, so the changes confidently can be attributed to HBO₂. This change correlates with the clinicians' reported stabilization of ORN.

HBO₂ is not generally used as the sole medical treatment of ORN. In fact, the use of HBO₂ in combination with surgery as a medical intervention for all types of ORN is still controversial.²⁸⁻³³ However, the findings of this study support the thesis that HBO₂ has a positive physiologic and psychological impact on some factors for this patient group.

References

- Office of National Statistics: Cancer Statistics Registrations: Registrations of Cancer Diagnosed in 2005, England. London, Office of National Statistics, 2008
- Chambers MS, Garden AS, Kies MS, et al: Radiation-induced xerostomia in patients with head and neck cancer: Pathogenesis, impact on quality of life, and management. *Head Neck* 26:796, 2004
- Chandu A, Smith AC, Rogers SN: Health-related quality of life in oral cancer: A review. *J Oral Maxillofac Surg* 64:495, 2006
- Hancock PJ, Epstein JB, Sadler GR: Oral and dental management related to radiation therapy for head and neck cancer. *J Can Dent Assoc* 69:585, 2003
- O'Sullivan B, Gullane P, Irish J, et al: Preoperative radiotherapy for adult head and neck soft tissue sarcoma: Assessment of wound complication rates and cancer outcome in a prospective series. *World J Surg* 27:875, 2003
- Clarke RE, Tenorio LM, Hussey JR, et al: Hyperbaric oxygen treatment of chronic refractory radiation proctitis: A randomized and controlled double-blind crossover trial with long-term follow-up. *Int J Radiat Oncol Biol Phys* 72:134, 2008
- Davis JC, Dunn JM, Gates GA, et al: Hyperbaric oxygen. A new adjunct in the management of radiation necrosis. *Arch Otolaryngol* 105:58, 1979
- Farmer JC Jr, Shelton DL, Angelillo JD, et al: Treatment of radiation-induced tissue injury by hyperbaric oxygen. *Ann Otol Rhinol Laryngol* 87:707, 1978
- Ferguson BJ, Hudson WR, Farmer JC Jr: Hyperbaric oxygen therapy for laryngeal radionecrosis. *Ann Otol Rhinol Laryngol* 96:1, 1987
- Marx RE: Radiation injury to tissue. *In* Kindwall EP (ed): *Hyperbaric Medicine Practice* (ed 2). Flagstaff, AZ, Best Publishing, 1995, pp 483-495
- Feldmeier JJ, Hampson NB: A systematic review of the literature reporting the application of hyperbaric oxygen prevention and treatment of delayed radiation injuries: An evidence based approach. *Undersea Hyperb Med* 29:4, 2002
- Feldmeier JJ: Hyperbaric oxygen for delayed radiation injuries. *Undersea Hyperb Med* 31:133, 2004
- Marx RE, Ames JR: The use of hyperbaric oxygen therapy in bony reconstruction of the irradiated and tissue-deficient patient. *J Oral Maxillofac Surg* 40:412, 1982
- Marx RE, Johnson RP, Kline SN: Prevention of osteoradionecrosis: A randomized prospective clinical trial of hyperbaric oxygen versus penicillin. *J Am Dent Assoc* 111:49, 1985
- Gerlach NL, Burkhuisen R, Kuanders JH, et al: The effect of hyperbaric oxygen therapy on quality of life in oral and oropharyngeal cancer patients treated with radiotherapy. *Int J Oral Maxillofac Surg* 37:255, 2008
- Harding SA, Hodder SC, Courtney DJ, et al: Impact of perioperative hyperbaric oxygen therapy on the quality of life of maxillofacial patients who undergo surgery in irradiated fields. *Int J Oral Maxillofac Surg* 37:617, 2008
- Ware JE Jr, Snow KK, Kosinski M, et al: SF-36 Health Survey. Manual and Interpretation Guide (ed 2). Lincoln, RI, Quality Metric, 2002
- Bjorndal K, Ahlner-Elmqvist M, Tolleson E, et al: Development of a European Organization for Research and Treatment of Cancer (EORTC) questionnaire module to be used in quality of life assessments in head and neck cancer patients. EORTC Quality of Life Study Group. *Acta Oncol* 33:879, 1994
- Bjorndal K, et al: A prospective study of quality of life in head and neck cancer patients. Part II: Longitudinal data. *Laryngoscope* 111:1440, 2001
- Clarke D: Effective patient blinding during hyperbaric trials. *Undersea Hyperb Med* 36:13, 2009
- Vissink A, Raghoobar GM, Roodenburg JL, et al: Ahlner-Elmqvist M, Hammerlid E: Does hyperbaric oxygen therapy improve quality of life? *Int J Oral Maxillofac Surg* 38:99, 2009
- Tabachnick BG, Fidell LS: Review of univariate and bivariate statistics. *In* Tabachnick BG, Fidell LS (eds): *Using Multivariate Statistics* (ed 5). Boston, Pearson, 2007, pp 33-59
- Reuther T, Schuster T, Mende U, et al: Osteoradionecrosis of the jaws as a side effect of radiotherapy of head and neck tumour patients—A report of a thirty year retrospective review. *Int J Oral Maxillofac Surg* 32:289, 2003
- Kluth EV, Jain PR, Stuchell RN, et al: A study of factors contributing to the development of osteoradionecrosis of the jaws. *J Prosthet Dent* 59:194, 1988
- Goldwasser BR, Chuang SK, Kaban LB, et al: Risk factor assessment for the development of osteoradionecrosis. *J Oral Maxillofac Surg* 65:2311, 2007
- Ware JE Jr: Standards for validating health measures: Definition and content. *J Chronic Dis* 40:473, 1987
- Hammerlid E, Silander E, Hörnemark I, et al: Health-related quality of life three years after diagnosis of head and neck cancer—A longitudinal study. *Head Neck* 23:113, 2001
- Annan D, Depondt J, Aubert P, et al: Hyperbaric oxygen therapy for radionecrosis of the jaw: A randomized, placebo-

- controlled, double-blind trial from the ORN96 study group. *J Clin Oncol* 22:4893, 2004
29. D'Souza J, Goru J, Goru S, et al: The influence of hyperbaric oxygen on the outcome of patients treated for osteoradionecrosis: 8 Year study. *Int J Oral Maxillofac Surg* 36:783, 2007
 30. Feldmeier JJ, Heimbach RD, Davolt DA, et al: Hyperbaric oxygen in the treatment of delayed radiation injuries of the extremities. *Undersea Hyperb Med* 27:15, 2000
 31. Granström G: Placement of dental implants in irradiated bone: The case for using hyperbaric oxygen. *J Oral Maxillofac Surg* 64:812, 2006
 32. Myers RA, Marx RE: Use of hyperbaric oxygen in post radiation head and neck surgery. *NCI Monogr*, 1990:151
 33. Sulaiman F, Hwryn JM, Zlotolow IM: Dental extractions in the irradiated head and neck patient: A retrospective analysis of Memorial Sloan-Kettering Cancer Center protocols, criteria, and end results. *J Oral Maxillofac Surg* 61:1123, 2003

4.3.4 Study Limitations

The major limitation to this study was the sample size. Everybody who fitted the criteria over the four years of recruitment and was treated at DDRC had their data included. To this extent, the study was of the whole population, rather than a sample. Even so, this rate of referral would require more than eleven years to obtain data from 50 people, which is what calculations suggest would be needed to allow for an understanding of variables such as age and gender, and even greater numbers would be required to look at treatment variable such as Gray of radiotherapy, time since treatment, location and staging of tumour etc.

4.3.5 Contribution to knowledge and autoethnographic issues

4.3.5.1 Contribution to knowledge

At the time of writing, paper 5 remains the only peer reviewed paper to be published, that specifically investigates the impact of HBOT on the QoL of people with Type III ORN. It also provides data that is directly comparable to people receiving HBOT for the side effects of radiotherapy for HNC, where HBOT is being used prophylactically.

Appendix 12 has an overview of how the research undertaken in paper 5 was disseminated prior to the paper's publication. It also provides a list of papers where paper 5 has been cited, and this is followed by a quality appraisal of the paper.

4.3.5.2 Autoethnographic issues

Working with this group of people made me aware that having a diagnosis of cancer might be traumatic, but living with the aftermath might be even worse. One problem with the types of delayed medical side effects I was seeing was their rarity. About four percent of HNC patients who receive radiotherapy as their treatment or part thereof develop ORN. Less than 1 percent develop type III ORN. I found myself questioning if I would readily agree to radiotherapy as part of my treatment if I had a cancer diagnosis, given the potential dreadful side effects and the experiences that had been reported to me. But as they told me; *'if I knew I was going to be part of the 1 percent that got this, then I would have said no. But what is one percent when your life is on the line'* [DM, 2004].

4.3.6 Informing the research journey

Patient experience is central to evaluating quality of care in the NHS and Department of Health. Although there are no national mandates or guidelines, there is a drive towards delivering services that patients want. There is also growing evidence to support the systematic use of patient reported outcome measures (PROMs) in routine clinical practice in cancer (Dimopoulos et al 2009). Quality of Life tools fit under the umbrella of PROMS and indeed holistic needs assessments. Benefits of collecting PROM data from patients include; early detection of physical and psychological symptoms that may otherwise be overlooked, monitoring disease progression, provision of information on the impact of prescribed treatment, facilitation of patient-clinician communication, and promotion of a model of shared decision making (Valderas et al 2008). In the management of chronic illness and cancer, PROMs have demonstrated improved processes and patient satisfaction with their care (Marshall et al 2006; Howell et al 2015). Following on from the research reported in papers 4 and 5, DDRC undertook to collect this data routinely and assess it on an individual basis to address the needs of patients whilst with the centre. This data needs collating and assessing as a cohort thereby providing a larger sample than that reported in either paper 4 or 5. It is hoped that this data will be analysed and reported after 100 people have completed the measures at all time points.

A similar phenomenon to type III ORN is Bisphosphonate induced necrosis of the mandible. Bisphosphonate-associated osteonecrosis of the jaw is an important condition seen most commonly in oncology patients receiving high-dose intravenous bisphosphonates. Low-dose bisphosphonates given either orally or intravenously in osteoporosis patients may also be linked to the development of osteonecrosis of the jaw. Some evidence has been forthcoming supporting the use of HBOT in combination with antibiotic treatments. However, more data would be useful, and there have been a number of cases treated at DDRC which would make a valuable case series. As mentioned previously in this chapter, embedding research into everyday practice would probably have meant that this data would have already been written up for dissemination at conference and for publication. Unfortunately, this has yet to happen at the DDRC, although progress is being made in that direction.

4.4 Summary

All the papers (3-5) look at change over the duration of a treatment (PR, paper 3; HBOT papers 4 & 5). They collect data from people before they undertake a treatment and then at the end, just before they are discharged from that episode of care. In this respect, they all share a clear common characteristic, making them ideal for cohort studies.

There are challenges with this type of design, and these come to the fore when collecting QoL data. The Hospital Anxiety and Depression Scale was used in all the papers in this chapter. In each case, it was given to people to complete at some point during the initial assessment that ensured their suitability for the intervention. This timing may capture people when they have heightened levels of anxiety due to the assessment process, rather than their ongoing feelings about their medical condition. Issues also arise when collecting data at the end of treatment/intervention. At this point people may have formed friendships with other patients, a rapport with staff, and be generally more comfortable with the expectations of the intervention. All of these may have an impact on reducing anxiety and depression and improving the other aspects of QoL being measured. It is also possible that people will unconsciously respond in a more positive way in order to unconsciously reward the clinical team for their care, and to maintain good relationships with those people that may have an impact on their future treatments.

Single sample prospective cohort studies, will always be criticised for being unable to control for these types of factors, with the critical call to include some form of control or control group. None of the three papers presented in this chapter used a control group. The reasons included; that as a charity DDRC, outside of the NHS, it does not have access to people who would be suitable for the intervention, but cannot access it without a referral from their NHS based care team. This is often going to be the case in pragmatic and embedded studies undertaken in clinical practice.

4.4.1 Summary of contribution to knowledge

Paper 3 disseminated information on the LINQ, the first tool designed with patients to assess their information needs rather than their knowledge. This enables clinicians to identify areas requiring particular attention, during routine clinical practice.

Papers 4 and 5 build on the evidence base of psychosocial impact of HBOT on people treated for HNC, thereby allowing clinicians to include these factors in their considerations as to whether to refer their patients for this treatment.

4.4.2 Summary of the autoethnographic issues

Undertaking this research reported in this chapter has highlighted the importance of listening to patients. The experiences showed me how vital it is to embed the research into the clinical setting, and that listening to patients ensures that the questions being asked are relevant and meaningful to improving patient care.

Lastly, all the people I spoke to during the data collection for papers 4 and 5, told about how finishing treatment did not equate to them reaching the end of their disease journey. How, being told about side-effects of treatments and the risk of experiencing them only becomes relevant when you become one of the 1%. It was brought home to me how varied an experience of cancer treatment can be, and how the ongoing experiences of these people are mostly overlooked.

Chapter 5 Longitudinal Studies

This chapter discusses longitudinal (multiple time points) studies. Longitudinal data provides us with an understanding of how a phenomenon changes over time. This is a vital piece of evidence that is often not available to people who develop interventions. Single and two time point studies can identify the prevalence of a phenomenon, but without longitudinal studies, a change attributed to an intervention could simply be a result of time passing. This type of study builds on single time point prevalence research and two time point (cross-sectional) cohort work.

This chapter reports on one paper covering primary research that investigates the longitudinal pattern of positive psychological change (PPC) in a head and neck cancer (HNC) population. This paper is built directly on the work presented in paper 2, Chapter 3.

5.1 Paper 6 - Clinical experience leading to the research work and production of the paper

It became clear very early on in my work with people who had had HNC that life did not return to normal a matter of months following completion of treatment.

Although it was vital to undertake the work presented in paper 2, there was an obvious absence of data that looked at the persistence, if any, of PPC.

My experience with cross-sectional research reported in chapter 4, and reviewing the literature of PPC made it clear to me that a piece of research that was truly longitudinal was needed. I chose to collect 5 years of data to understand a person's journey over the duration of their contact with a hospital for their cancer. That is to say, from at least 3 months post treatment to 5 years or more when they would have been formally discharged from the cancer service.

5.1.1 Research Questions

After the completion of treatment for Head and Neck Cancer, what are the trajectories for positive psychological change longitudinally (defined as longer than 12 months)?

5.1.2 Study Design

Two commonly used designs for studies examining PPC are the cross-sectional design and the longitudinal design. The cross-sequential design represents an alternative to these designs, which aims to correct some of the problems inherent in the cross-sectional and longitudinal designs. Farrington (1991) states that the cross-sequential strategy is *“a way of achieving the benefits of the longitudinal method while minimizing the problems”* (p. 369). The specific advantages are that it allows intra-individual changes to be assessed within a shorter follow-up period than that required for a traditional longitudinal design and that the use of multiple cohorts increases confidence in the generalisability of the results to the sample population as a whole (Miller, 2007).

It is important to be mindful of the limitations of the research methodology design, and the extent to which legitimate and accurate conclusions can be drawn. I considered that this was the appropriate method to undertake data collection for the current work, due to the timeframe available, the nature of the trauma (cancer), and the desire to understand a longitudinal pattern of development and change. It could be argued that a traditional longitudinal design may be methodologically stronger; however it is confounded by the time of measurement. It would also require more resources and a longer time frame than I had available. Good research design is always a balance between the research question being addressed, resources available, and the particular circumstances of the study population.

Traditionally, cross-sequential designs recruit people for a set period of time and have overlapping cohorts. Due to the possibility of the HNC cohort having a recurrence or dying, this process was adapted in the research, with the different cohorts being constructed. Each year the previous cohort potential participants were reviewed to ensure they still met the inclusion criteria. If they did, even if they did not respond the previous year, they were sent a cover letter and a copy of the survey. Then the medical records of all the new potential participants were reviewed and where appropriate sent a set of questionnaires, thereby constructing a modified new cohort year on year.

The strength of this method is its ability to compare the development (longitudinally) of PPC and comparison of cohorts. This study design has the advantage of being able to unpick factors such as whether age or birth cohort makes a difference.

The two key disadvantages of this study design that were overcome were; its complex nature and that it is time consuming to run because of the continued annual review of data and ongoing recruitment. The findings are arguably still only generalisable to cohorts and the historical periods of time measured.

The same data collection methodology as paper 2 (Section 3.2.2) was used in this work, with the inclusion of new participants year on year to improve the granularity of the data, and allow for a larger sample size over an extended time period.

5.1.3 The trajectory of positive psychological change in a head and neck cancer population

Research Paper
 Head and Neck Oncology

The trajectory of positive psychological change in a head and neck cancer population

Harding
 Department of Health and Social Sciences,
 University of the West of England, Bristol, UK

S.A. Harding: The trajectory of positive psychological change in a head and neck cancer population. Int. J. Oral Maxillofac. Surg. 2017; xxx: xxx–xxx. © 2017 International Association of Oral and Maxillofacial Surgeons. Published by Elsevier Ltd. All rights reserved.

Abstract. A stressful event may be sufficient to challenge a strongly held set of assumptions about the world and the self. In some people this may lead to post-traumatic stress disorder (PTSD) and in others to positive psychological change (PPC), whereby a person's reactions to the challenge are beneficial. Little research has investigated PPC in people who have had head and neck cancer (HNC). The aim of this study was to identify demographic, clinical, and psychological factors associated with PPC over time. A cross-sequential study collected data over 5 years. Participants were sent the Silver Lining Questionnaire (SLQ; a measure of PPC), the University of Washington HNC quality of life measure, and the Medical Outcomes Short-Form 12 each year. Additional data were collected from clinical records. Analysis using linear mixed-effects modelling revealed that participants with lower stage tumours and those who only had a surgical intervention reported greater PPC over time. Multivariable modelling adjusting for psychosocial variables found that PPC had a quadratic relationship with time since diagnosis, increasing initially and levelling off after 18 months. These findings build on the minimal PPC research with people following HNC. In particular it demonstrates a model of trajectories for the development of PPC longitudinally over time.

Key words: positive psychological change; post-traumatic growth; longitudinal.

Accepted for publication 20 September 2017

A cancer diagnosis is a highly stressful event and may be significant enough to shatter an individual's understanding of themselves. In 1975 Parkes used the phrase 'assumptive world' to refer to a way in which a person views their reality¹. According to Parkes, we are rarely aware of the fundamental elements of our assumptive world; the minor disappointments, challenges, and failures of day-

to-day life seldom bring them to light. It has been said that they are conservative cognitive schemas that resist change and disconfirmation². The questioning of the basic assumptions is what fractures the assumptive world and triggers the rebuilding of them to accommodate new realities². Sometimes this may lead to people developing post-traumatic stress disorder (PTSD), but it has also been shown that

people can positively re-evaluate aspects of their lives^{3–8}.

There is a growing body of literature supporting positive psychological change (PPC) following a range of traumas, including natural disasters, bereavement, and illness^{9,10}. However, to date there has been no single term used consistently in the literature. In 1991, Yalom and Lieberman used the term 'positive psychological

2 Harding

changes', but this has also been referred to as 'perceived benefits', 'benefit finding', 'thriving', 'stress-related growth', 'adversarial growth', 'post-traumatic growth' (PTG), and 'existential growth'¹¹. Although varying underlying theories have been proposed and their authors have chosen specific terms, Tedeschi and Calhoun suggest that they are often direct synonyms, or include significant elements of, PPC¹². Scales developed to assess PPC from these different theoretical standpoints have been investigated and appear to measure an umbrella concept that can be labelled PPC. They may vary in the nature of the specific subscales that can also be reported¹³.

At the time of writing, the vast majority (68%) of quantitative research into PPC involving people with cancer has focused on breast cancer¹⁴. The pervasiveness of breast cancer research is likely due to the prevalence of the disease, the size of the population that it affects, and the availability of funding to investigate the impact of the disease and its treatment. However, the make-up of that population is different from those people diagnosed with head and neck cancer (HNC), with more than 99% of breast cancer patients being female. The equivalent figure is less than 45% in people with HNC. Higher levels of economic deprivation appear to have a greater role in HNC than in breast cancer¹⁵. To date, only six studies and one systematic review have been published that focus on PPC in HNC^{16–22}.

Four of the six HNC PPC articles identified used a cross-sectional study design^{17–20}; the other two used a prospective design with two time-points, the second time-point being only 6 months after baseline^{21,22}. The most common methodology of data collection in non-HNC PPC studies has been a single time-point or cross-sectional method^{23–25}. However, with cancer therapies becoming more effective, people are living longer following treatment. It is therefore important to understand the longitudinal pattern of PPC development, so that it may be understood in relation to coping and the future development of rehabilitation services.

Helgeson et al. undertook a meta-analytic review that included a mixture of trauma cohorts²⁶. These cohorts came from backgrounds that included natural disasters, bereavement, and illness. In studies in which the time since the traumatic event was more than 2 years, they found that PPC was related to lower levels of depression and a more positive effect, whereas PPC was related to higher levels

of global distress when time since the traumatic event was less than 2 years. Helgeson et al. also found that PPC was related to a reduction in anxiety when the time since the traumatic event was 2 years or less²⁶. This suggests that, as time elapses, PPC is more likely to reflect significant life changes and/or reprioritization of life values in response to the trauma rather than coping mechanisms.

All of the longitudinal studies that have investigated possible trajectories of PPC within a cancer cohort have so far focused on people who have previously been diagnosed as having breast cancer^{25,27–30}. Danhauer found that those who reported elevated levels of PPC at 4 months post diagnosis maintained these levels and could be described as having early-onset PPC²⁹. They also found that, in those people who reported early-onset PPC, higher levels of PPC were found in proportion to increasing time from diagnosis.

The aim of the current study was to investigate the pattern or trajectory of development of PPC within an HNC population and how this changes over time. A secondary area of investigation was to explore how biological, social, and psychological variables are associated with PPC.

Materials and methods

Design

This study used a 5-year, cross-sequential design with self-completion psychometric measures.

Procedure

Ethical review was sought and granted. Potential participants were identified through a regional health-informatics database. Questionnaire materials were sent to all potential participants annually each October. No follow-up letters were sent. Data collection was undertaken over a duration of 5 years. All potential participants (those matching the inclusion criteria, and not previously excluded) were approached through the Head and Neck Directorate of the regional hospital leading this study. As people with newly diagnosed HNC reached the inclusion criteria (being 3 months post-treatment), they were added to the list of people sent the questionnaire materials. A time frame of >3 months post-treatment was selected to allow for the acute effects of treatment to resolve and the demands of treatment (e.g. fatigue, travel, financial burden, family upheaval) to have lessened.

All questionnaires were sent out as hard copy via the Royal Mail, along with a stamped, return-addressed envelope. People who in previous years had indicated that they did not wish to answer the questionnaires were not included in subsequent years.

Participants

To be deemed eligible, a person had to be over the age of 18 years, with an understanding of English judged by clinical staff to be sufficient to complete a series of questionnaires in English. Tumours had to have a histological diagnosis of squamous cell carcinoma (SCC) and be sited in the mouth, lip, oral cavity, salivary gland, pharynx, nasal cavity, or sinuses.

People were not approached, or were excluded from one or more rounds of data collection, if they were newly diagnosed with cancer (less than 3 months post-treatment) or had a tumour or recurrence in their HNC or in a location not included in this research.

A total of 416 completed or partially completed questionnaire materials were returned by 185 people over the 5 years of data collection. Table 1 shows the number of questionnaires sent and returned by year of recruitment. Demographic data collected included age at time of diagnosis, gender, Index of Multiple Deprivation (IMD)³¹, and family status (married, living with partner, living alone, living with relative/friends). Medical data collected included tumour stage, date of diagnosis, treatment regimen, and date of treatment completion. Treatment regimen was split into three categories: surgery, surgery and radiotherapy, and radiotherapy (with or without chemotherapy) but without surgery.

No significant differences in demographic and medical data were found between the responders and non-responders to the questionnaires (Mann-Whitney *U*-test). Table 2 provides demographic information on the respondents in relation to time since treatment completed.

Table 1. Number of questionnaire sets sent out and returned across the 5 years of data collection.

	Sent	Returned (%)
Year 1	151	61 (44)
Year 2	166	74 (45)
Year 3	194	95 (49)
Year 4	211	95 (45)
Year 5	239	85 (36)

Table 2. Biological and social characteristics by time since treatment completed.

	Time since treatment completed, months							P-value
	3-6	7-12	13-18	19-24	25-36	37-60	≥61	
IMD, mean ± SD	19.02 ± 11.82 (n = 37)	18.73 ± 9.56 (n = 33)	20.91 ± 10.52 (n = 22)	26.78 ± 13.65 (n = 11)	23.39 ± 12.53 (n = 20)	19.43 ± 8.63 (n = 23)	19.43 ± 8.63 (n = 25)	0.362
Gender, n								
Male	26	26	15	8	13	17	18	
Female	14	11	7	3	7	6	7	
Age at diagnosis, years, mean ± SD	65.59 ± 11.54 (n = 35)	63.43 ± 8.93 (n = 37)	59.41 ± 9.05 (n = 22)	59.55 ± 12.91 (n = 11)	64.95 ± 15.34 (n = 20)	58.87 ± 10.86 (n = 23)	57.64 ± 10.69 (n = 25)	0.042
Family status, n								
Married/living with partner	26	24	13	7	16	14	17	
Living alone	9	6	4	3	1	5	5	
Living with relatives/friends	2	0	0	1	2	1	1	
Cancer stage, n								
1	9	5	3	2	4	8	6	
2	5	2	3	4	4	5	7	
3	5	11	3	0	2	5	8	
4	19	15	9	3	9	5	4	
Treatment regimen, n								
Surgery	16	11	5	5	8	10	9	
Surgery + RT	14	14	12	2	7	8	14	
RT with or without CT (no surgery)	8	12	4	4	4	5	2	

IMD, Index of Multiple Deprivation; SD, standard deviation; RT, radiotherapy; CT, chemotherapy. n = number of participants completing the measure.

Materials

The Silver Lining Questionnaire (SLQ), University of Washington Quality of Life Questionnaire version 4 (UW-QOL), and Medical Outcomes Short-Form 12 (SF-12) were the questionnaires used.

The SLQ is a 38-item measure that examines the extent to which people believe their illness has led to PPC despite the negative consequences of being ill³².

The UW-QOL, which is specific for HNC, has 12 domains: pain, appearance, activity, recreation, swallowing, chewing, speech, shoulder function, taste, saliva, mood, and anxiety, and includes two sub-scales of physical and social function³³. UW-QOL has been validated by comparison to the Karnofsky scale and Sickness Impact Profile, demonstrating an average criterion validity of 0.85³³.

The SF-12 is a generic, health-related quality-of-life questionnaire with 12 items³⁴. Results for each patient are expressed in terms of two meta-scores: physical component summary (PCS) and mental component summary (MCS).

Analysis

Linear mixed-effects models (LMM) were used to assess the effects of demographic, medical, and psychological variables on the SLQ. Random coefficient models were used to assess the effects of these variables on the SLQ across all seven time-points. The models recognize the existence of data hierarchies, allowing for residual components at each level in the hierarchy^{35,36}. Time was calculated as months since diagnosis and was included in the model using both linear and quadratic terms. The intercept and time slope were included as random effects in the models. Predictor variables included in the model were demographic (age at time of diagnosis, gender, family status, IMD), medical (tumour stage), and psychological (UW-QOL and SF-12) variables.

The analysis was undertaken using IBM SPSS Statistics version 23.0 (IBM Corp., Armonk, NY, USA) and the LMM function³⁷, and following the process prescribed by Leech et al.³⁵. IBM SPSS uses REML (restricted maximum likelihood) as the estimation method. This adjusts for the fixed effects, usually leading to a reduction in standard error. It is the default approach in IBM SPSS.

Results

Data for 'time since treatment completion' were collected as interval data and

4 Harding

grouped into categories guided by the clinical practice. Analysis of variance (ANOVA) with post-hoc analysis indicated that the SLQ did not show any significant change over time. No subscale of health-related quality of life (as measured by the SF-12) or HNC-specific quality of life (as measured by the UW-QOL) factors changed over the seven time-points. Age at time of diagnosis showed a significant difference on ANOVA, but post-hoc Tukey analysis failed to locate the time points where a significant difference lay. This is because the Tukey procedure controls the type 1 error rate and requires a larger difference to declare significance compared to when no adjustment is used.

Medical factors associated with PPC up to 11 years post treatment

The LMM indicated that stage of the tumour and the treatment regimen both had a significant effect on reported PPC ($P = 0.009$ and $P = 0.003$, respectively) over time. Further analysis with ANOVA and post-hoc testing indicated that over the duration of data collection, participants with stage I tumours at time of diagnosis reported more PPC than those

with higher stage tumours (3 or 4) at time of diagnosis ($F = 1.533$, $P = 0.027$). Multivariable longitudinal modelling with 3–12-month data as a baseline supported this finding (estimate 3.433, $P = 0.009$).

Over the same time period, participants who had surgery alone reported more positive change than both those who had surgery with radiotherapy ($t = 2.317$, $P = 0.021$) and those who were not treated surgically, but who had received radiotherapy with or without chemotherapy ($t = 3.213$, $P = 0.001$). In the multivariable model that adjusts for psychosocial variables, SLQ had a quadratic relationship with time since diagnosis, increasing initially after diagnosis and levelling off over time (Fig. 1). The multivariable baseline model also supported this finding, indicating surgery as the only treatment regimen that was significantly associated with higher SLQ scores (estimate 3.997, $P = 0.003$).

Psychosocial factors associated with PPC up to 11 years post treatment

When modelled using LMM, the SLQ did not show any association with gender or family status ($P = 0.053$ and $P = 0.366$,

respectively). Socio-economic status, as measured by the IMD, was associated with PPC with the SLQ. This indicated that those participants with high or low socio-economic status reported greater levels of PPC than those in the middle of the scale.

Linear mixed-effects modelling using the total score of the UW-QOL found an association with positive change ($P = 0.009$), but due to the redundant parameters on the subscales, no significant associations were found with individual subscales. The longitudinal model with the SLQ as dependent variable failed to find any relationship with the SF-12 physical or mental component scores.

Psychosocial factors associated with PPC between baseline and longitudinal time points

LMM were undertaken on the difference between the baseline data (3–12 months) and the longitudinal time points. The medical and psychosocial variables were fixed factors within the model and therefore were not included as predictors in the model.

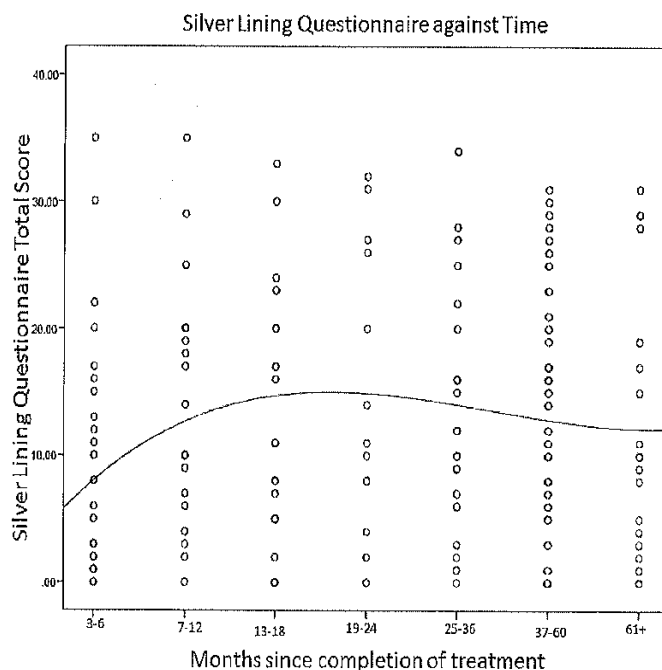


Fig. 1. Quadratic relationship between Silver Lining Questionnaire results and months since completion of treatment.

Please cite this article in press as: Harding SA. The trajectory of positive psychological change in a head and neck cancer population. *Int J Oral Maxillofac Surg* (2017), <https://doi.org/10.1016/j.ijom.2017.09.010>

The difference between baseline data and longitudinal data was modelled using LMM with SLQ as the dependent variable. Only the PCS of the SF-12 and seven subscales of the UW-QOL (chewing, taste, activity, recreation, swallowing, saliva, and mood) showed that an improvement in these health-related measures over time was associated with the generation of PPC in the participants.

Discussion

A notable strength of this research is the use of a cross-sequential design. This methodology corrects for some of the problems inherent in the cross-sectional design frequently used in this field of study and also for the expense of longitudinal designs. Farrington states that the cross-sequential strategy is "a way of achieving the benefits of the longitudinal method while minimizing the problems"³⁸ (p. 369). The specific advantages are that it allows intra-individual changes to be assessed within a shorter follow-up period than is required for a traditional longitudinal design. Its use of multiple cohorts increases confidence in the generalizability of the results to the sample population as a whole³⁹.

The comparatively low incidence of HNC in the wider population of cancer patients makes it challenging to undertake longitudinal studies. Despite good return rates, the relatively small population of potential participants led to a small sample size in this study. One of the major strengths of LMM is that it allows for data to be included even if a person does not respond at all time points. Despite this, the subscales of the UW-QOL and SF-12 were found to have been redundant in the LMM analysis.

In modelling the longitudinal nature of PPC within a HNC population, the stage of the tumour and the treatment regimen were both found to have a relationship with the total score of the SLQ. The participants with lower stage tumours and those who only had a surgical intervention reported more PPC. In the multi-variable model that adjusts for psychosocial variables, SLQ had a quadratic relationship with time since diagnosis, increasing initially after diagnosis and levelling off after about 18 months. This pattern of findings is a development from the research of Harding and Moss¹⁷, in which participants with low stage tumours (stage 1) reported a higher level of PPC than those with stage 2 and 3 tumours and noticeably higher than those with stage 4 tumours at 3–12 months post-treatment.

However the treatment patterns were the same over both the short and long term¹⁷.

The SLQ did not show any longitudinal association when modelled with gender or family status. Other social factors did show a relationship with PPC; participants with high or low socio-economic status as measured by the IMD reported greater levels of PPC than those participants in the middle of the scale. When the data were compared at baseline (between 3 and 12 months) and at other data points, stage of tumour, treatment regimen, and socio-economic status were significantly associated with higher SLQ scores. The modelling of baseline and longitudinal data with the SLQ as the dependent variable found that the PCS subscale of the SF-12 and seven subscales of the UW-QOL (chewing, taste, activity, recreation, swallowing, saliva, and mood) were associated with higher levels of PPC. This suggests that there is a change in the characteristics of PPC and associated variables after the initial 3–12 months post treatment. It is conceivable that in the short time frame PPC is more related to coping or short-term benefit-finding than the development of a more permanent change in perspective. The analysis suggests that in the longer term, the PCS subscale of the SF-12 and the UW-QOL may be the more meaningful variables associated with the development or sustaining of PPC.

It is clear from the current study that further research is needed to gain greater insight into the bio-psychosocial factors relating to the development of PPC. Further investigation may also elucidate whether multiple trajectories can be differentiated within a HNC population in much the same way as those present in a breast cancer population²⁹. However, it must be acknowledged that this cancer occurs in a smaller proportion of the population than others such as breast cancer¹⁵, and may therefore require multi-centre collaboration to achieve sufficient statistical power.

A driving force of the current study was to address one of the noteworthy problems with the measurement of PPC – the lack of longitudinal studies – whilst noting the continued problem of a lack of baseline data collected prior to the events. Jayawickreme and Blackie have suggested that participants must undertake five steps for each item on a PPC questionnaire: (i) deduce current standing on the factor or dimension being asked about, (ii) recall prior standing on the dimension before the event had occurred, (iii) compare these standings (i ii), (iv) calculate the degree of change, and finally (v) evaluate how

much of the change was due to the traumatic event⁴⁰. These steps highlight the debate about whether developed PPC scales actually measure change. There is little option but to rely on a respondent's retrospective self-assessment of change when investigating a historical event within a temporal framework. These challenges are acknowledged, but it is believed that the use of PPC measures is an appropriate way to gain an understanding of a cohort's development of PPC, as is the use of similar measures in PTSD research. However, it must be recognized that focused qualitative research may provide insight into the social, contextual, and environmental factors that may influence the development and progression of PPC.

Tedeschi and Calhoun have promulgated a model of post-traumatic growth that indicates that positive change can co-exist with PTSD symptoms, as distress is necessary to engender the processes considered central to PPC⁹. They have reported that different domains of functioning may be affected differently by trauma, with some domains positively affected and others being harmed. It is possible that survivors will experience distress related to trauma at the same time as they experience PPC. An example of this may be where a person is experiencing fear of recurrence due to being sent a review appointment letter, but is grateful that family members are able to offer comfort and support. In the same vein as Tedeschi and Calhoun⁹, Joseph et al.⁴¹ have suggested that trauma may leave a person sadder, yet with an enhanced appreciation of what is important and a greater commitment to live in accordance with these values. A person reporting 'new-found meaning and clarity of life priorities' would still be considered to have experienced a positive change, even if that person is not reporting feeling more satisfied with life. The ability to experience both PPC and PTSD simultaneously is a phenomenon that needs further investigation and understanding if healthcare professionals are to enhance their interactions with people following the diagnosis and treatment of HNC.

Beyond an understanding of the development of PPC, it should be incumbent on healthcare professionals and service users to harness its potential as an intervention or as an element of an intervention package. A meta-analysis assessed the relationship between intervention participation and PTG, but failed to find any studies that included an outcome measure of PPC⁴². Roepke has suggested that there is a modest increase in PPC following

6 Harding

intervention⁴², but due to the limited research reported on the natural development and time course of PPC, it is possible that even this modest increase could be due to the passage of time. Future clinical practice needs to be mindful of these factors and, in order to assess their effectiveness, needs to include a measure of PPC in the development and delivery of interventions.

At the time of writing, UK guidelines on the psychological management of HNC patients after treatment focused on managing psychological distress and social support, both of the person who had HNC and their family⁴³. These guidelines recognize that the majority of people appear to cope, but there are people who experience negative psychological effects, such as fear of recurrence, changes to their daily lives, and sense of self⁴³. In the current set of guidelines there is no mention of PPC, which is unsurprising given the need to offer intervention and treatment to those people with psychological problems. If people are doing well, there is little imperative for the National Health Service (NHS, UK) to engage with them or provide services for them.

As noted by Calhoun and Tedeschi⁴⁴ (p. 13), "posttraumatic growth is common, but it is by no means universal". It is clear from the results of the present study and previous research focusing on PTSD that not all people who have been treated for HNC experience PPC or PTSD; however, clinicians should be aware of both these possible outcomes and the range between them.

Funding

This project was self-funded by the lead author.

Competing Interests

None.

Ethical approval

Ethical approval was sought and received by proportional review via the Integrated Research Application System (IRAS) (167514 – 15/EM/0052).

Patient consent

Not required.

Acknowledgments. Dr Tim Moss is acknowledged for his supervision of the

author's doctorate and Mr Simon Williams for his review of the manuscript.

References

1. Parkes CM. What becomes of redundant world models? A contribution to the study of adaptation to change. *Br J Med Psychol* 1975;48:131–7. <http://dx.doi.org/10.1111/j.2014-8341.1975.tb02315.x>.
2. Janoff-Bulman R, Schwartzberg S. Toward a general model of personal change. In: Snyder CR, Forsyth DR, editors. *Handbook of social and clinical psychology: the health perspective*. New York: Pergamon Press; 1991. p. 488–508.
3. Collins RL, Taylor SE, Skokan LA. A better world or a shattered vision? Changes in life perspectives following victimization. *Soc Cogn* 1990;8:263–85. <http://dx.doi.org/10.1521/soco.1990.8.3.263>.
4. Giovenco G, McDougall J. Logotherapy: a journey into meaning for people with AIDS. *Int Janna Logother* 1994;17:76–81.
5. Laerum E, Johnsen N, Smith P, Larsen S. Myocardial infarction may induce positive changes in life-style and in the quality of life. *Scand J Prim Health Care* 1988;6:67–71. <http://dx.doi.org/10.3109/02813438809009293>.
6. LaFortune-Fredette SL. Breast cancer survivors: concerns and coping. *Cancer Nurs* 1995;18:35–46.
7. O'Connor AP, Wicker CA, Germino RB. Understanding the cancer patient's search for meaning. *Cancer Nurs* 1990;13:167–75.
8. Taylor SE. Adjustment to threatening events. A theory of cognitive adaptation. *Am Psychol* 1983;38:1161–73. <http://dx.doi.org/10.1037/0003-066X.38.11.1161>.
9. Tedeschi RG, Calhoun LG. Posttraumatic growth: conceptual foundations and empirical evidence. *Psychol Inq* 2004;15:1–18. http://dx.doi.org/10.1207/s15327965psi1501_01.
10. Updegraff JA, Taylor SE. From vulnerability to growth: positive and negative effects of stressful life events. In: Harvey J, Miller F, editors. *Loss and trauma: general and close relationship perspectives*. Philadelphia, PA: Brunner-Routledge; 2000. p. 3–28.
11. Yalom ID, Lieberman MA. Bereavement and heightened existential awareness. *Psychiatry* 1991;54:334–45. <http://dx.doi.org/10.1080/0033272.1991.11024563>.
12. Tedeschi RG, Calhoun LG. Posttraumatic growth. In: Friedman HS, editor. *Encyclopedia of mental health*. Second edition. Waltham, MA: Academic Press; 2016. p. 305–7.
13. Joseph S, Linley A, Harris GJ. Understanding positive change following trauma and adversity: structural clarification. *J Loss Trauma* 2004;10:83–96. <http://dx.doi.org/10.1080/15325020490890741>.
14. Casellas-Grau A, Ochoa C, Ruini C. Psychological and clinical correlates of posttraumatic growth in cancer: A systematic and critical review. *Psychooncology* 2017. <http://dx.doi.org/10.1002/pon.4426>. [Epub ahead of print].
15. Cancer Research UK. Oral cancer statistics. <http://www.cancerresearchuk.org/health-professional/cancer-statistics/statistics-by-cancer-type/oral-cancer> [Accessibility verified June 28, 2017].
16. Harding S, Sanipour F, Moss T. Existence of benefit finding and posttraumatic growth in people treated for head and neck cancer: a systematic review. *PeerJ* 2014;11:e256. <http://dx.doi.org/10.7717/peerj.256>.
17. Harding S, Moss T. The impact of treatment for head and neck cancer on positive psychological change within a year of completing treatment. *Int J Oral Maxillofac Surg* 2017. <http://dx.doi.org/10.1016/j.ijom.2017.07.023>. [Epub ahead of print].
18. Harrington S, McGurk M, Llewellyn CD. Positive consequences of head and neck cancer: key correlates of finding benefit. *J Psychosoc Oncol* 2008;26:43–62. <http://dx.doi.org/10.1080/07347330802115848>.
19. Ho S, Rajandram RK, Chan N, Samman N, McGrath C, Zwahlen RA. The roles of hope and optimism on posttraumatic growth in oral cavity cancer patients. *Oral Oncol* 2011;47:121–4. <http://dx.doi.org/10.1016/j.oraloncology.2010.11.015>.
20. Holtmaat K, van der Spek N, Cuijpers P, Leemans CR, Verdonck-de Leeuw IM. Post-traumatic growth among head and neck cancer survivors with psychological distress. *Psychooncology* 2016;26:96–101. <http://dx.doi.org/10.1002/pon.4106>.
21. Leong AM, Nik Jaafar NR, Zakaria H, Rajandram RK, Mahadevan R, Mohamad Yunus MR, Shah SA. Posttraumatic growth, depression and anxiety in head and neck cancer patients: examining their patterns and correlations in a prospective study. *Psychooncology* 2015;24:894–900. <http://dx.doi.org/10.1002/pon.3740>.
22. Llewellyn CD, Horney DJ, McGurk M, Weinman J, Herold J, Altman K, Smith FF. Assessing the psychological predictors of benefit finding in patients with head and neck cancer. *Psychooncology* 2013;22:97–105. <http://dx.doi.org/10.1002/pon.2065>.
23. Bellizzi KM, Smith AW, Reeves RB, Alfano CM, Bernstein L, Meeske K, Baumgartner KB, Ballard-Barbash RR. Posttraumatic growth and health-related quality of life in a racially diverse cohort of breast cancer survivors. *J Health Psychol* 2010;15:615–26. <http://dx.doi.org/10.1177/1359105309356364>.
24. Tomich PL, Helgeson VS, Nowak Vache EJ. Perceived growth and decline following breast cancer: a comparison to age-matched controls 5 years later. *Psychooncology* 2005;14:1018–29. <http://dx.doi.org/10.1002/pon.914>.
25. Wang AW, Chang CS, Chen ST, Chen DR, Hsu WY. Identification of posttraumatic growth trajectories in the first year after

Please cite this article in press as: Harding SA. The trajectory of positive psychological change in a head and neck cancer population, *Int J Oral Maxillofac Surg* (2017), <https://doi.org/10.1016/j.ijom.2017.09.010>

- breast cancer surgery. *Psychooncology* 2014;23:1399–405. <http://dx.doi.org/10.1007/s00431-014-2577-7>.
26. Helgeson VS, Reynolds KA, Tonich PL. A meta-analytic review of benefit finding and growth. *J Consult Clin Psychol* 2006;74:797–816. <http://dx.doi.org/10.1037/0022-006X.74.5.797>.
 27. Danhauser SC, Case LD, Tedeschi R, Russell G, Vishnevsky T, Triplett K, Ip EH, Avis NE. Predictors of posttraumatic growth in women with breast cancer. *Psychooncology* 2013;22:2676–83. <http://dx.doi.org/10.1007/s00431-013-2298-2>.
 28. Danhauser SC, Crawford SL, Farmer DF, Avis NE. A longitudinal investigation of coping strategies and quality of life among younger women with breast cancer. *J Behav Med* 2009;32:371–9. <http://dx.doi.org/10.1007/s10865-009-9211-x>.
 29. Danhauser SC, Russell G, Case LD, Sohl SJ, Tedeschi RG, Addington EL, Triplett K, Van Zee KJ, Nattalis EZ, Levine B, Avis NE. Trajectories of posttraumatic growth and associated characteristics in women with breast cancer. *Ann Behav Med* 2015;49:650–9. <http://dx.doi.org/10.1007/s12160-015-9696-1>.
 30. McDonough MH, Sabiston CM, Wrosch C. Predicting changes in posttraumatic growth and subjective well-being among breast cancer survivors: the role of social support and stress. *Psychooncology* 2014;23:114–20. <http://dx.doi.org/10.1007/s00431-014-2330-0>.
 31. Department for Communities and Local Government. The English indices of deprivation 2007. <http://www.communities.gov.uk> [Accessibility verified].
 32. Sodergren SC, Hyland ME. What are the positive consequences of illness? *Psychol Health* 2000;15:85–97. <http://dx.doi.org/10.1080/08870440008400290>.
 33. Rogers SN, Gwanne S, Lowe D, Humphris G, Yuchi B, Weymuller Jr FA. The addition of mood and anxiety domains to the University of Washington quality of life scale. *Head Neck* 2002;24:521–9. <http://dx.doi.org/10.1002/hed.10106>.
 34. Ware Jr J, Kosinski M, Keller SD. A 12-item short-form health survey: construction of scales and preliminary tests of reliability and validity. *Med Care* 1996;34:220–33.
 35. Leech NI, Barrett KC, Morgan GA. *SPSS for intermediate statistics: use and interpretation*. Third edition. London, UK: Lawrence Erlbaum Associates; 2008.
 36. West BT, Welch KB, Galecki AT. *Linear mixed models: a practical guide using statistical software*. Second edition. London, UK: Chapman and Hall/CRC Press; 2014.
 37. SPSS Inc. Linear mixed-effects modeling in SPSS: an introduction to the mixed procedure. SPSS Technical Report, 2005. <https://gou.glj97Mmi> [Accessibility verified June 28, 2017].
 38. Farrington DP. Longitudinal research strategies: advantages, problems, and prospects. *J Am Acad Child Adolesc Psychiatry* 1991;30:369–74. <http://dx.doi.org/10.1097/00004583-199105000-00003>.
 39. Miller SA. *Developmental research methods*. Third edition. New York: Sage Publications; 2007.
 40. Jayawickreme E, Blackie LE. Post-traumatic growth as positive personality change: evidence, controversies and future directions. *Eur J Pers* 2014;28:312–31. <http://dx.doi.org/10.1002/per.1963>.
 41. Joseph S, Linley PA, Andrews L, Harris G, Howle B, Woodward C, Shevlin M. Assessing positive and negative changes in the aftermath of adversity: psychometric evaluation of the changes in outlook questionnaire. *Psychol Assess* 2005;17:70–80. <http://dx.doi.org/10.1037/1040-3590.17.1.70>.
 42. Roepke AM. Psychosocial interventions and posttraumatic growth: a meta-analysis. *J Consult Clin Psychol* 2015;83:129–42. <http://dx.doi.org/10.1037/a0036872>.
 43. Humphris G. Psychological management for head and neck cancer patients: United Kingdom National Multidisciplinary Guidelines. *J Laryngol Otol* 2016;130(S2):S45–8. <http://dx.doi.org/10.1017/S0022215116000426>.
 44. Calhoun LG, Tedeschi R. *Posttraumatic growth in clinical practice*. Mahwah, NJ: Routledge; 2013.

Address:
 Sam Harding
 Department of Health and Social Sciences
 University of the West of England
 Bristol
 BS16 1QY
 UK
 Tel.: +44 7944 363973
 E-mail: sharding.jb@gmail.com

5.1.4 Study Strengths and Limitations

5.1.4.1 Study Design

Research into PPC and cancer has used a number of different baseline definitions e.g.: 1) post diagnosis, to pre treatment, 2) post surgery, but while chemo, radiotherapy may still be on-going, 3) immediately following treatment. It is my contention that any change in PPC, measured in a cross-sectional study, using these baseline time points, would always find a positive change. How could a person not report feeling better than they did at the time they were diagnosed with cancer, or during their treatment?

It could, however, be argued that the research undertaken for both papers 2 and 6 does not have a baseline measurement, as no data was collected before diagnosis or at least before treatment. The theory of PPC requires there to have been a traumatic or seismic event in a person's life, for their assumptions to have been shattered. It is not practicable to attempt to administer the full suite of research measures on everyone at each general practitioner's appointment on the off chance that it might be the appointment that precedes a diagnosis of cancer. The question then becomes when to measure.

Most people who receive a diagnosis of cancer are referred to hospitals for investigation of their symptoms. During these investigations and likely at the time of referral for investigation, they will be told that cancer is a possible diagnosis, and it is likely that they will have elevated levels of anxiety. Measuring PPC at this point would be misleading, as a diagnosis has not been given and confirmation of the traumatic event is still pending. The negative effect of elevated anxiety and worry at this time would mean that a return to normal levels of psychological functioning may appear to a researcher to be PPC. The same problems could be seen if the initial measurement was undertaken post-diagnosis, but pre-treatment as it was by Danhauer *et al.* (2009) with a breast cancer cohort, Steele *et al* (2008) with people with Hepatobiliary cancer and Llewellyn *et al.* (2013) with a similar HNC cohort to the current research. Silva *et al.* (2012) added in further variation where the first measurement time point was post-surgery, the second time point was 'during adjuvant' treatment of chemotherapy, radiotherapy or chemo-radiotherapy, and at their third time point 28% of their respondents were still having hormonotherapy,

even though the authors say that this last measurement was 6 months after treatment had been completed.

This variation in choice of time point for data collection requires consideration to be given to the nature of cancer; is it a chronic or acute event? It can be argued that cancer is a chronic extreme event in clinical and conceptual terms. However, some people may see it as an acute event due to a single short treatment, such as a surgical intervention, with a good known outcome (Holman and Lorig, 2000). I would argue that cancer should not be regarded as an acute trauma, but, rather, that it should be considered a chronic stressor due to;

- the difficulty in identifying a sole stressor,
- the internal nature of the illness,
- the temporal orientation of the person's expectations of the future as well as memories of the diagnosis,
- treatments and other challenges that may be experienced due to having HNC,
- the practical impossibility of establishing the onset and termination of the traumatic event, and,
- differences in perceived control.

Measuring PPC while treatment is on-going is likely to be while the person is still experiencing extreme stress. This may be why the majority of peer review journal articles report the time of data collection either in relation to time since diagnosis or time since treatment completion.

Time since diagnosis is a commonly used and interesting point of reference (Bellizzi *et al.*, 2010; Danhauer *et al.*, 2015; Leong *et al.*, 2015; Manne *et al.*, 2004). It provides an absolute reference point which would be the same for all participants, but it does not allow for variation in the time individual treatment regimens can take and the subsequent variation in time since completion of treatment that would be present within a participant pool.

The other time point model used is the time since treatment had been completed. The time chosen varies between studies without any consensus, Pat-Horenczyk *et al.* (2015) required people to be at least 3 months post treatment, while Sears *et al.* (2003) measured PPC at 20 weeks post-treatment. Harrington *et al.* (2008)

undertook a prospective study and HNC participants were between 6 months and 10 years post-treatment. While Holtmaat *et al.* (2016) only recruited people that were at least 1 month post-treatment, and Ho *et al.* (2011) required people to have completed therapy for at least 6 months.

Consideration of the impact of the different time frames on change in PPC, accounting for the various and continuous stressors experienced by people with cancer, and to have a consistent reference point for all people completing the measures, were all deliberated during the construction of the study leading to papers 2 and 6.

A difference between the research undertaken leading to papers 2 and 6 and other research in this area is that this work sent out the measures to all appropriate potential participants during the first week in October for five consecutive years. Other researchers have had participants' complete measures at specific times, such as 20 weeks post-treatment. The method used in this study has the benefit of providing a greater resolution of data. Collecting data at a single calendar time point means that some participants will be just over 3 months post-completion of treatment, and some will be nearly 12 months post-completion. Using this method, if a sufficient sample size could be collected; it would be possible to have a data point for each week post-treatment. The annual nature of the data collection did mean that the individual respondents were only approached once, and therefore minimised the burden on them to return multiple surveys within a year. This is a noticeable difference from those studies that collect multiple time points over a relatively short duration, such as Wang *et al.* (2014) who collected data one day, and then 3, 6, and 12 months following surgery.

5.1.4.2 Participant response rates

The data collection methodology meant that there is missing information. The phenomenon of missing data is commonly referred to as data holes. After five years of data collection, survey returns stood at a total of 416 from 185 patients and;

- 71 people out of a potential 289 returned the survey pack once
- 53 people out of a potential 230 returned the survey pack twice
- 24 people out of a potential 178 returned the survey pack three times
- 15 people out of a potential 146 returned the survey pack four times

- 22 people out of a potential 57 returned the survey pack five times

Not all of these returns could be used in the analysis due to incomplete responses. According to the research design, only the data from those people that returned the survey pack three or more times (n=61) were included in the longitudinal analysis. The use of linear mixed-effects models meant that even with data holes all 61 people's data would be included in the analysis.

In relation to response rates, this equated to between 36 and 49 percent of questionnaires being returned. This is in line with the literature which suggests that, for the length of survey used in the current study (65 questions), a 40% return rate is common (Iglesias *et al.*, 2002). As mentioned in section 3.2.4.2 the scales used in this research had completion rates between 48 and 95 percent. Some of the reasons for incomplete responses are also presented in chapter 3, with discussion of the validity of the selected measures.

A challenge with this research is the relatively small participant pool. Similar research with a breast cancer cohort recruited more than 600 women covering an 18 month data collection (Danhauer, 2015). With this sample size, they were able to identify multiple trajectories, whereas I only found one within my research. If a multi-centre study could be funded it would be interesting to see if more than one trajectory could be identified within a HNC population.

5.1.5 Contribution to knowledge and autoethnographic issues

5.1.5.1 Contribution to knowledge

The research leading to paper 6 has created new knowledge through the identification and characterisation of a trajectory in the development of PPC in a HNC population. Trajectories have previously been identified in a breast cancer population (Danhauer, 2015), but not with HNC. It has further contributed to knowledge by covering a longer time span, over 5 years post treatment. This represents the first longitudinal study into PPC within a HNC population, where previous research has stopped at 12 or 18 months following diagnosis or treatment.

This research contributed further through the use of a cross-sequential methodology in a HNC population. The adoption of this cross-sequential methodology allowed for longitudinal data collection in a population that present challenges for long term data collection due to the nature of the HNC and patterns of survival. Demonstrating the

appropriateness and advantages of cross-sequential methodologies in examining PPC, encourages others to adopt this methodology in future.

Appendix 13 has an overview of how the research undertaken in paper 6 was disseminated prior to the paper's publication. It also provides a list of papers where paper 6 has been cited, and this is followed by a quality appraisal of the paper.

5.1.5.2 Autoethnographic issues

The paper referenced in this chapter represents one of the most complex pieces of research I have initiated. It was conducted over an extended period of time (over 5 years) meaning I had to establish clear protocols and processes that would survive the extended period. As this work was self-funded (except postage costs, covered by the hospital) I did not have the luxury of a large research team. I needed to work smart to have the time and psychological resources to maintain and build my relationship with my PI group. This provided an internal governance check by my PI members. This relationship allowed me to ensure the research processes and interpretation remained relevant to the patient cohort.

The cross-sequential design has been under-utilised because it is perceived to be complex; I needed to ensure that I could maintain a focus on the fundamental research questions without getting diverted by methodological complications. The analysis also involved complex statistical tools that I had not used before. For the analysis phase, I worked closely with a statistician to ensure that I chose the right tools, and deployed them properly.

One of the major frustrations for me as a researcher is the grant application game. Papers 2 (chapter 3) and 6 were undertaken by me self-funding the studies. My frustrations are not driven by the process, but by the realisation that if a person is working in a specialised area, even if they can show their investigation would lead to significant patient impact, they are still less likely to receive funding than those working with a larger disease group, e.g. HNC vs breast cancer. This has made me more cynical, and more determined to move my research interests forward.

Another aspect of the research game is the need for team. I find myself talking to junior researchers and explaining that although it is up to them to drive 'their' project they are not on their own. They are important but research is a 'Team Sport' (Figure 3).

5.1.6 Future research questions

In modelling the longitudinal nature of PPC, the stage of the tumour and the treatment regimen both have a relationship with the total score of the Silver Lining Questionnaire (SLQ). The participants with lower stage tumours and those who only had surgical intervention report more PPC.

In the multivariable model that adjusts for psychosocial variables, SLQ had a quadratic relationship with time since diagnosis, increasing initially after diagnosis and levelling off over time, after about 18 months. Longitudinally the SLQ did not show any association when modelled with gender or family status. Other social factors did show a relationship with PPC. Participants with high or low socio-economic status as measured by the IMD reported greater levels of PPC than those participants in the middle of the scale. This differs from the results of the short-term baseline data reported in paper 2.

Positive change, with fewer associated problems, was impacted most by chewing, speech and taste from the University of Washington (UoW) questionnaire, suggesting how these elements of eating were important to the respondents. The longitudinal model with the SLQ as dependent variable did not find a relationship with the Medical Outcomes Short Form-12 (SF-12) or any of its sub-scales.

In reviewing the literature around PPC I was led to question if this was a real phenomenon, or an illusion, a subjective experience, rather than an objective change (Sumalla *et al.*, 2009; Zoellner and Maercker, 2006). There is the possibility that reported PPC masked a lack of coping and/or denial. Another possibility is that it is used as an acute coping strategy. If PPC is a long term coping strategy, this could explain the seeming contradiction of a person's ability to show PPC whilst also reportedly experiencing post-traumatic stress disorder (PTSD) symptoms. Future studies could look at this contradiction within an HNC cohort.

I was once again left with a number of questions. Are there mediators and moderators for the development of PPC? Do patients' and clinicians' perspectives differ in their views of the severity of the cancer as a traumatic event, and if it is an acute or chronic experience? Can interventions stimulate PPC in a HNC population? These questions are all considered in appendix 15.

Figure 3: There is an I in team



But even here there is frustration. Having an idea, wanting to lead on the investigation and then having it taken by more senior research staff is upsetting. This is especially the case when there is the expectation that I will still be the one actually undertaking the research. How these situations are managed is an on-going part of my development. I am therefore very upfront with the researchers who I am advising/guiding, and seek to make all my interactions collaborative.

5.2 Summary

5.2.1 Summary of contribution to knowledge

The research presented within paper 6 created new knowledge both methodologically and conceptually by the identification and characterisation of trajectories in the development of PPC in a HNC population.

5.2.2 Summary of autoethnographic issues

There are two main impacts of undertaking the research reported in papers 2 and 6 (both part of the same longitudinal project). First, the importance of maintaining and building meaningful, trusting and honest relationships with people. Here, I am talking about the people who have experienced HNC and their commitment to making my research better by ensuring it is relevant to themselves and those who will have HNC in the future. Second, recognising the pleasure of working with a team. This team will of course be in part represented by the Public Patient Involvement (PPI) group formed, but it goes wider, such as the ability to work collaboratively in the day-to-day undertaking of research tasks, and figuring out ways to embed those tasks in clinical routine. Facilitating research to happen pragmatically, so that the benefits of positive outcomes can be rapidly picked up by other settings and used to improve patient care and pathways.

I feel very strongly, that research must be disseminated. Traditional peer reviewed journal articles can spread your findings widely, but only if they are read. Word of mouth from clinical teams potentially provides more credibility within service providers, and the more buy-in from the clinical team, the more they are likely to talk about it to colleagues. My hope is that this will lead to quicker uptake within practice, and encourage more health care professionals to embed research into their daily lives.

Chapter 6 Systematic Reviews

This chapter presents three systematic reviews. All were undertaken to examine the current state of research knowledge in their area of interest. They differ in their reasons for being undertaken, and what the authors present to the readers. The first paper (7) examined the state of literature in order to collate the quantitative data and look at patterns of biopsychosocial factors. The second paper (8) looked at differences in the characteristics of two groups when studied in naturalistic environments. The third paper (9) is a sub-set of a larger review. It takes papers identified as being relevant to preschool children's speech and language and refines the retained papers to those related to speech sound disorders (SSD), and then further categorizes them by intervention. Each review was undertaken following the PRISMA guidelines, and reported using the PRISMA flowchart.

6.1 Paper 7 - Clinical experience leading to the research work and production of the paper

This systematic review is part of the corpus reported in papers two and six, looking at positive psychological change (PPC) present in people following treatment for head and neck cancer (HNC). I needed to select the measures to administer to potential participants, and understand the findings of previous research that had used these measures, and the cohorts with whom they had been used. During this initial work I found a surprisingly large number of scales reported as measuring positive change (Appendix 16). It was clear that there was a growing body of literature supporting the suggestion that a stressful or traumatic event may be a catalyst for PPC (Tedeschi and Calhoun, 2004; Updegraff and Taylor, 2000). However, as with the range of measures, there has been no single term used consistently in the literature. In 1991, Yalom and Lieberman used the term 'positive psychological changes'. These positive changes, have also been referred to as 'perceived benefits', 'benefit finding', 'thriving', 'stress-related growth', 'adversarial growth', 'post-traumatic growth', or 'existential growth', and were written about in light of changes in the perceptions of oneself, social relationships with family and friends and life priorities and appreciation of life.

The term 'Post-traumatic growth' is now the most widely used term due to its ability to describe the need for individuals to have experienced trauma before they

experience positive change over time. As previously noted in this thesis and the previous two papers (2 and 6), the term PPC has also been used. The choice of PPC over post-traumatic growth was made due to the nature of the trauma experienced by the participants in this research. In presenting work on PPC to people who have received a diagnosis of HNC the researcher has found that the word 'growth' has significant negative meaning, as it is a word associated with a cancerous tumour (Section 3.2.6). In working with this group of people, it became evident that the phrase positive psychological change was better received and facilitated communication with the researcher.

At the time of undertaking this review, I was still new to this area of study and trying to define my own understanding of the concept. I was having numerous conversations with academics, clinicians and people who had experienced HNC. I was forming the opinion that all of the described phenomenon could all be covered by the umbrella term of PPC. However, an early set of reviewer's comments on paper 2, made it very clear that this reviewer saw Benefit Finding and Posttraumatic Growth as distinctly different. I felt I needed to reconfigure the systematic review to present positive change in the short term as Benefit Finding and in the long term as Posttraumatic Growth, although now I feel I would probably argue a stronger case for both Benefit Finding and Posttraumatic Growth being subsets of the larger concept of PPC.

6.1.1 Research Questions

This systematic review (paper 7) aimed to collate the current (search date: February, 2012) quantitative data to understand how differing medical, psychological and social characteristics of HNC may lead to Benefit Finding/Posttraumatic Growth.

6.1.2 Study Design

The review strategy was a minor adaption of the Cochrane Collaboration systematic review methodology (e.g., we did not use the EMEZ search strategy) and uses a narrative synthesis (The Cochrane Collaboration, 1999) and guidance from Petticrew & Roberts (2006). Booth and Fry-Smith's (2004) PICO model (Population, Intervention, Comparison, Outcome) guided the development of the search strategy.

Once the PICO model was framed and search strategy designed, five separate searches were conducted in electronic databases; Pubmed, Psych Info (CSA), Psyc

Articles (CSA), OVID Medline, and Published International Literature on Traumatic Stress, to identify appropriate studies in articles published from the earliest entries of any of the databases until February 2012. No limits were placed on the electronic search in relation to age range of participants studied or language of publication.

All identified manuscripts were checked for inclusion by me and one of my co-authors, and then again we both checked the quality of the research using the Critical Appraisal Skills Programme Cohort Study appraisal tools (Critical Appraisal Skills Programme, 2011).

6.1.3 Existence of benefit finding and posttraumatic growth in people treated for head and neck cancer: a systematic review

Existence of benefit finding and posttraumatic growth in people treated for head and neck cancer: a systematic review

Sam Harding¹, Fatimeh Sanipour¹ and Timothy Moss²

¹ Department of Psychology, University of the West of England, Bristol, United Kingdom

² Centre for Appearance Research, University of the West of England, Bristol, United Kingdom

ABSTRACT

Background. The impact of head and neck cancer (HNC) in long-term survivors differs widely among individuals, and a significant number of them suffer from the negative effects of disease, whereas others report significant positive effect. This systematic review investigated the evidence the implications of treatment for HNC and subsequent development of Benefit Finding (BF) or Posttraumatic Growth (PTG).

Purpose. To understand how differing medical, psychological and social characteristics of HNC may lead to BF/PTG and subsequently inform post-treatment interventions to encourage positive outcomes.

Method. In February 2012, five databases including Pubmed, and Psych Info, were searched, for peer-reviewed English-language publications. Search strings included key words pertaining to HNC, BF, and PTG. One thousand three hundred and sixty three publications were identified, reviewed, and reduced following Cochrane guidelines and inclusion/exclusion criteria specified by a group of maxillofacial consultants and psychologists. Publications were then quality assessed using the CASP Cohort Critical Appraisal tool.

Findings. Five manuscripts met the search and selection criteria, and were sourced for review. All studies were identified as being level IIb evidence which is a medium level of quality. The majority of studies investigated benefit finding (80%) and were split between recruiting participant via cancer clinics and postal survey. They focused on the medical, psychological and social characteristics of the patient following completion of treatment for HNC.

Conclusion. Demographic factors across the papers showed similar patterns of relationships across BF and PTG; that higher education/qualification and cohabitation/marriage are associated with increased BF/PTG. Similarly, overlap with disease characteristics and psychosocial factors where hope and optimism were both positively correlated with increased reported BF/PTG.

Subjects Oncology, Psychiatry and Psychology

Keywords Posttraumatic growth, Benefit finding, Head and neck, Cancer, Silver lining questionnaire, Posttraumatic growth inventory, Quality of life, Systematic review

Submitted 9 October 2013

Accepted 12 January 2014

Published 11 February 2014

Corresponding author

Sam Harding, sharding.jb@gmail.com

Academic editor

Gerhard Andersson

Additional Information and
Declarations can be found on
page 18

DOI 10.7717/peerj.256

© Copyright

2014 Harding et al.

Distributed under
Creative-Commons CC-BY 3.0

OPEN ACCESS

How to cite this article Harding et al. (2014), Existence of benefit finding and posttraumatic growth in people treated for head and neck cancer: a systematic review. PeerJ 2:e256; DOI 10.7717/peerj.256

INTRODUCTION

A great deal of evidence has accumulated over the past thirty years for the negative sequelae of trauma. Traumatic events can include a range of experiences including health threats. The literature on coping with health difficulties has documented a variety of negative consequences, including; depression (e.g., *Moyer & Salovey, 1996; Timberlake et al., 1997*), posttraumatic stress disorder (PTSD) (e.g., *Alter et al., 1996; Andrykowski et al., 1998*), and adjustment difficulties (e.g., *Schulz et al., 1995*). These models have tended to work towards a clinical diagnosis for which treatment may be prescribed.

By contrast, models of positive illness recovery have been informed by a range of more general theories of life change (*Horowitz, 1986; Park & Ai, 2006; Paton, 2006*). These have tried to understand the mechanisms that may underpin the positive sequelae of health-related trauma. Since these models are not working towards a diagnosis for prescription, there has been no imperative to coalesce around a common agreed understanding against which a diagnosis can be made.

Morse (1997) conceptualises coping with life-threatening illness as incorporating five distinct stages. The first stage is uncertainty or vigilance, during which patients suspect a condition and attempt to maintain emotional control whilst trying to understand their condition and its severity. The second stage is disruption, a time when individuals realise that they are affected by what they perceived to be a serious disease and may experience high levels of stress. In the third stage, striving for recovery, individuals may try to gain control over their illness with the help of personal and environmental resources. The fourth stage is striving to restore one's self and making sense of altered reality. The fifth and final stage is learning to live with the altered self, in which patients attain a new equilibrium as a result of accepting the illness and its consequences. In chronic illness, a return to a prior state of health may not be a realistic outcome. This and subsequent models suggest that it is the time of diagnosis, and the disruption stage, especially when this involves news of a life-threatening illness, that patients are the most likely to experience trauma (*Morse & Johnson, 1991*). This is also the stage during which individuals are most likely to confront existential issues posed by the diagnosis (*Doka, 2008*).

Brennan (2001) proposes that social cognitive transition (SCT) model builds on previous theories of coping, traumatic stress, social-cognition and cognitive theories of emotion. This theory hinges on the central components of the cognitive models of PTSD, except it allows for both positive and negative psychological outcomes after a trauma. *Brennan (2001)* proposes that all individuals have mental models of the world, made up of assumptions. As an individual interacts with their world these assumptions are either confirmed or disconfirmed by experience. If we consider Leventhal's model of Self-regulation (*Leventhal, Nerenz & Steele, 1984*), then his stimulus is a disruption or challenge to the Assumptive World. The arising representations map to an expectation, and the coping behaviours to new experiences. The subsequent outcomes either confirm or disconfirm the mental model of the Assumptive World. In this way, Brennan's medical

model encompasses Leventhal's broader psychosocial framework and provides an account for the diverse psychosocial outcomes experienced by cancer patients.

This model would propose that PTSD is the negative result of an extremely troubling event that is highly incongruent with the individual's assumptions about the world. Brennan (2001) indicates that denial and avoidance are the first responses of a traumatised individual, which create more stress and potentially lead to the development of new assumptions about the world, assumptions that may be dysfunctional and lead to heightened levels of distress or PTSD. However, avoidance and denial can also serve a positive roll by diluting "the absorption of 'traumatic' information" (Brennan & Moynihan, 2004, p. 9). Conversely, Brennan & Moynihan (2004) proposes that an adaptive response to traumatic experiences requires worry. It is hypothesised that worry is a part of the cognitive attempt to anticipate and prepare for future threat (Brennan & Moynihan, 2004; Eysenck, 1992). By imagining and confronting worst case scenarios, by "decatastrophising" them, the individual can appraise the realistic nature of the event. Brennan & Moynihan (2004) proposes that positive outcomes from traumatic experiences can then occur, as unrealistic goals or outcomes are discarded and implicit long-standing life goals become clear and distinct.

Benefit finding (BF) and posttraumatic growth (PTG) describe similar outcomes following adversity, yet there are clear differences. Both describe a positive outcome with BF being described as the acquisition of benefit from adversity (Collins, Taylor & Skokan, 1990; Tennen & Affleck, 2002) and PTG growth being the success with which individuals coping with the aftermath of trauma reconstruct or strengthen their perceptions of self, others and the meaning of events (Tedeschi & Calhoun, 1996). Examples of BF finding include a positive change in relationships, a greater appreciation of life and a change in life priorities. PTG is also described as 'the experience of significant positive change arising from the struggle with a major life crisis', with examples of increased sense of personal strength, changed priorities and richer existential and spiritual life being cited in the literature (Calhoun et al., 2000).

Despite these similarities, there is emerging evidence that there are critical differences, for example, Sears, Stanton & Danoff-Burg (2003) showed that BF was predicted by personal characteristics (i.e., education, optimism, and hope), but PTG was not. Benefit finding may start immediately after diagnosis and results from challenges to the individual's cognitive representations; that is, they have the same personal representations, but have positive ways of coping. By contrast, PTG is a re-assembly of the assumptive world in a new way following trauma and develops as a result of the rumination and restructuring of the self/world relationship, that occurs in the weeks, months, and even years following trauma and is focussed on changes in one's capacity to deal with adverse events (Calhoun & Tedeschi, 1998). So PTG results from challenges to deeper cognitive representations than BF and result in changed 'rules for living' and 'core schema', whereas BF may be more superficial and transient in nature. This difference may also lead one to expect more PTG growth with increasing time post-trauma, because more time is available for cognitive processing (Sears, Stanton & Danoff-Burg, 2003).

However, this hypothesis has yet to be tested and given that PTG has no diagnostic period of onset, unlike PTSD (*American Psychiatric Association, 2013*), this systematic review has aggregated BF and PTG and will search for both of these concepts and words/phrase used synonymously such as 'stress-related growth' and 'existential growth'. The authors will refer to these concepts throughout the remainder to this manuscript as BF/PTG unless making specific reference to information from research where one theoretical perspective has been purposely selected.

Recent studies have provided evidence that these positive processes also take place in chronically ill patients, including individuals suffering from cancer (*Affleck & Tennen, 1996; Carver & Antoni, 2004; Petrie et al., 1999; Schulz & Mohamed, 2004; Sears, Stanton & Danoff-Burg, 2003; Tomich & Helgeson, 2004*). The bulk of this research has been undertaken on females with breast cancer (*Carver & Antoni, 2004; Petrie et al., 1999; Sears, Stanton & Danoff-Burg, 2003; Tomich & Helgeson, 2004*). There have also been some general cancer review papers published, but none which have focused on people with head and neck cancer (*Stanton, Bower, & Low, 2006; Sumalla, Ochoa, & Blanco, 2009*). In the United Kingdom 125.9 females in every 100,000 will suffer from breast cancer and 1.0 males. For oral cancer the figures are 5.5 and 12.4 respectively (*Cancer Research UK, 2013*). Additionally *Cancer Research UK (2013)* statistics indicate that people with oral cancer are older at diagnosis than those with breast cancer. These two factors combined with the location of the tumour may impact the development of BF/PTG, and it is for this reason that a systematic review of this cancer site is needed.

This systematic review investigates the literature on BF/PTG in the patients treated for cancer in the region of the Head and Neck (HNC). The aim is to collate the current quantitative data to understand how differing medical, psychological and social characteristics of HNC may lead to BF/PTG and subsequently may inform diagnosis and future post-treatment interventions to encourage sustained positive outcomes.

METHODS

The review strategy was adapted from the Cochrane Collaboration systematic review methodology and uses a narrative synthesis (*The Cochrane Collaboration, 1999*) and guidance from *Petticrew & Roberts (2006)*.

Identification of selection criteria

The *Booth & Fry-Smith (2004)* PICO model (population, intervention, comparison, outcome) guided the development of the search strategy.

The 'Population' of interest was defined as adults (> 18 years) of either sex with HNC. Children and adolescents can develop HNC, but due to high relevance of developmental stage, and cognitive maturity they are excluded from the review. Terminal patients and those with recurrent metastatic disease on entry to the study were excluded, as they would currently be experiencing significant on-going challenging and potentially traumatic experiences.

This systematic review is not investigating an 'Intervention' in the sense of 'Cognitive Behavioural Therapy', as an example. The interventions of interest that may affect

Table 1 ICD10 codes related to cancer sites and incidence.

Cancer site	ICD10 code	Number of registrations 2000	Incidence: crude rate per 100,000, 2000	
			Men	Women
Mouth, lip & oral cavity	C00-06	2329	5.9	3.7
Salivary glands	C07-8	422	1	0.8
Pharynx	C09-14	1339	4	1.6
Nasal cavity, ear & sinuses	C30-31	352	0.8	0.6
arynx	C32	1903	6.6	1.3
Thyroid	C73	1131	1.3	3.3

outcome is the treatment for the malignant tumour, i.e., surgery, radiotherapy, chemotherapy and any combination of these treatments, or specifically named variations such as photodynamic therapy. In relation to 'comparisons', no limitations were put on the search strategy. However it was noted that comparison may be possible by simply comparing intervention groups, cancer sites (Table 1) or measure pre and post intervention.

When considering the relevance of 'outcome' measures to the development of the search strategy, this review focused purely on quantitative studies. The studies must include 'paper and pencil' or 'computer based' psychometrically sound measures of BF and/or PTG. This will allow comparison of statistical analysis of the relationship between BF/PTG and categorical medical and social variables, as well as other psychological characteristics collected via validated measures. Data collected via studies reporting qualitative data only were excluded.

Search strategy

The search strategy was designed in consultation with a senior librarian and the search terms following a review of the literature and discussion with a Maxillofacial Consultant (Supplemental Information A). A combination of 'free text' terms with Boolean operators and truncations were used. Five separate searches were conducted in electronic databases; Pubmed, Psych Info (CSA), Psyc Articles (CSA), OVID Medline, and PILOTS (Published International Literature on Traumatic Stress), to identify appropriate studies in articles published from the earliest entries of any of the databases until February 2012. No limits were placed on the electronic search in relation to age range of participants studied or language of publication. The PRISMA checklist was followed and a flow chart (Fig. 1) details the process of article selection.

The citations retrieved from each database were exported to 'Reference Manager 11' bibliographic management software (Thomson ResearchSoft, 2000). Duplicates were removed, and article screened for relevance, removing animal studies and medical and psychological studies which had been retrieved as they contained one or more of the search terms, e.g., Squamous Cell or Benefit (Supplemental Information B). To this point in the review process no limits or restrictions had been placed on 'cancer site' while

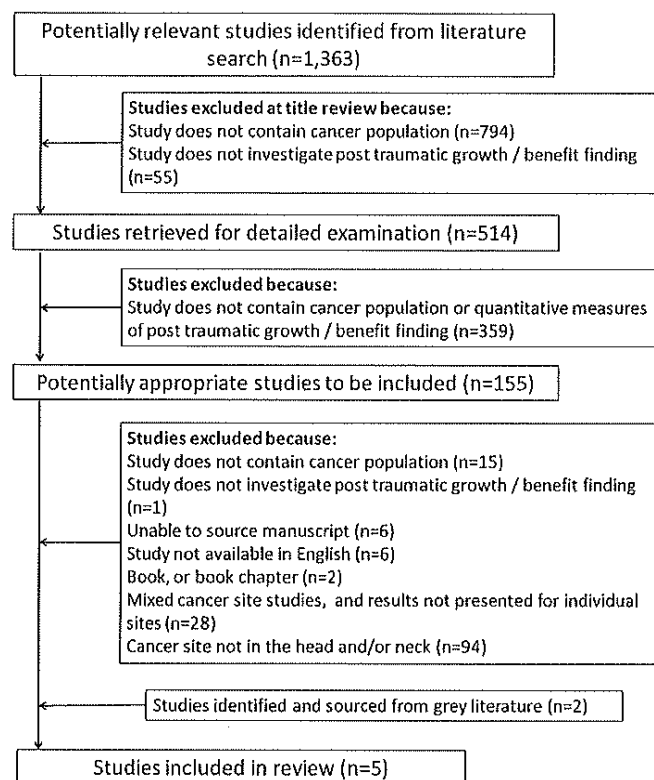


Figure 1 PRISMA flowchart.

searching the electronic databases or retrieved articles. This enabled papers reporting on multiple cancer sites to be identified and integrated for patterns between tumour locations. Supplemental Information B provides the list of search terms used to identify appropriate tumour locations within the head and neck region. We did not limit the search to include or exclude any type of intervention within this participant cohort. In this review, an intervention would be the type of cancer treatment they received. Cancer location and treatment were specific factors that were identified as potential confounders/variables within the selected papers, but this did not require additional terminology to be added to the research strings or strategies. The 514 abstracts of the remaining articles related to BF, PTG and/or cancer were screened by SH and twenty percent randomly sampled were reviewed by TM and FS.

Guidelines, dissertations and theses greater than 5 years old, handbooks, commentaries, review articles, expert opinions and case reports, as well as trials with fewer than ten participants were excluded, as were qualitative studies. Disagreement between the review authors was resolved by consensus through discussion. This identified

'potentially relevant articles' ($n = 155$) and these were obtained and appraised critically.

Three articles (Harrington, McGurk & Llewellyn, 2008; Ho et al., 2011; Llewellyn et al., 2011) were identified from this search strategy. After completing the literature search, references from these articles, review articles, thesis and books were examined to identify additional grey literature and the author (SH) contacted researchers identified. Two projects were identified, but no responses were received when the authors were contacted. Two of the authors of this Systematic Review (SH & TM), have two manuscripts in preparation for submission and these were included in this review as grey literature (S Harding & T Moss, 2013a, unpublished data; S Harding, T Moss, 2013b, unpublished data).

The five identified manuscripts were summarised separately, including a description of the study design, sample size, measurement, and time since diagnosis or treatment of HNC, and are presented in Tables 2 and 3.

One of the five identified papers did not provide sufficient data to extract as part of this review. The authors of that article were approached and subsequently provided an additional publication that enabled a fuller understanding of their data and greater comparison with other published work (Horney et al., 2011).

Quality assessment

This review has identified a very limited number of studies; it is therefore insufficient to limit the assessment of papers to those with the 'best' methodology. The studies identified in this review all represented 'level IIB' evidence (Supplemental Information C; *National Institute for Clinical Excellence*, 2004), or those at a medium level of quality, where high levels would refer to studies in the top of the hierarchy of evidence (e.g., systematic reviews, randomised controlled trials), and 'low' refers to those near the bottom of the hierarchy (case series, case reports, expert opinion). Given this assessment of quality, the remaining assessment of quality reflects variation within that small banding.

Quality was assessed using the Critical Appraisal Skills Programme (CASP) Cohort Study appraisal tools (*Critical Appraisal Skills Programme*, 2011). This tool provides a 12 point check list of study validity, risk of bias in recruitment, exposure, outcome measurement, confounding factors, reporting of results and the transferability of findings (maximum score of 12). The key questions from CASP were taken as a template for the quality appraisal (Supplemental Information D). The appraisal questions were answered with 'yes', 'can't tell' and 'no'. Where 'yes' was used, the study was felt to fill the criteria for that question. Where 'can't tell' was used, the study was considered to meet some of the criteria for the question, but not others. Where 'no' was used, the study was considered to explicitly not meet the criteria for the question. CASP does not provide cut-offs for quality levels, however no studies were ruled out on the basis of the quality appraisal since quality levels were similar between studies.

All identified manuscripts were checked for quality against the appraisal tool independently by SH and FS and confirmed by TM. Consensus was immediate between the reviewers. Each of the scales used within the studies were also assessed and reported

Table 2 Study descriptors.

Study	Author(s)	Aim of the study	Study design	Study measures	Demographic factors	Medical factors	Time of measurement
1	Harrington, McGurk & Llewellyn (2008)	(1) to determine the extent to which patient treated for HNC experience positive consequences of their illness, (2) to identify factors associated with benefit finding among this patient group	Cross-sectional postal survey	Benefit finding scale (BFS), Hospital Anxiety and Distress Scale (HADS), Life Orientation Test-Revised (LOT-R), Brief COPE	Age, Gender, Ethnicity, Education, Employment, Marital status	Type of treatment, time since last treatment, diagnosis of further illness since treatment, site, type of cancer and stage of cancer	0-6mths = 1, 6-12mths = 3, 13-24mths = 7, 25-47mths = 20, 48-72mths = 19, 73-121mths = 26
2	Llewellyn et al. (2011)	(1) to determine the extent to which patient treated for HNC experience positive consequences of their illness, (2) to establish the relationship between BF, other patient-reported outcomes and predictive factors such as coping strategy and level of optimism	Repeated measures prospective study using self-completion questionnaires	Benefit finding scale (BFS), Hospital Anxiety and Distress Scale (HADS), Life Orientation Test (LOT-R), Brief COPE, Medical Outcomes Short Form 12 (SF-12), Two-item measure derived from The European Organization for Research and Treatment (EORTC) of Cancer Quality of Life Questionnaire (QLQ-C30)	Age, Gender, Ethnicity, Education, Employment, Marital status	Type of treatment, site and stage of cancer	T1 = Between diagnosis and start of treatment, T2 = 6 months after completion of treatment
3	Ho et al. (2011)	Investigate if PTG occurs in oral cancer patients and if hope and optimism shows significant positive correlation with PTG	Cross-sectional postal survey	Chinese Posttraumatic Growth Inventory (PTGI), Hope scale (HS), Life Orientation Test - Revised (LOT-R)	Age, Gender, Religion, Education level, income	Time since diagnosis, stage of disease, and treatment type	Mean time was 3.6yrs (SD 0.34)
4	S Harding & T Moss, (2013a, unpublished data)	Investigate the relationship between BF, demographical, biomedical and HRQoL following the treatment for HNC	Cross-sectional postal survey	Silver Lining Questionnaire (SLQ), University of Washington Head and Neck Cancer Quality of Life (UoW), Medical Outcomes Short Form 12 (SF-12)	Age at diagnosis, Age at time of completing questionnaire, Gender, Ethnicity, Index of Multiple Deprivation, Occupation, Family Status	Tumour site, Stage of disease, Location of tumour, Treatment	Mean time from completing treatment to completing questionnaires 27.30mths (Range 3-76; SD 21.8)

5	S Harding & T Moss, (2013b, unpublished data)	Investigate the longitudinal relationship between BF, demographical, biomedical and HRQoL following the treatment for HNC	Repeated measures prospective cross-sectional study using self completion questionnaires	Silver Lining Questionnaire (SLQ), University of Washington Head and Neck Cancer Quality of Life (UoW), Medical Outcomes Short Form 12 (SF-12)	Age at diagnosis, Age at time of completing questionnaire, Gender, Ethnicity, Index of Multiple Deprivation, Occupation, Family Status	Tumour site, Stage of disease, Location of tumour, Treatment
---	---	---	--	--	--	--

(Supplemental Information E). Upon reviewing the studies' data collection tools and statistical analysis it became apparent that there was too great a variation between them and thus it was not appropriate to conduct additional analysis such as a meta-analysis using the reported findings.

RESULTS

Quality Assessment Findings

The fashion in which data is collected may affect the results. Two of the included studies collected the data during patients' clinic visits (*Ho et al., 2011; Llewellyn et al., 2011*). This may have increased the potential sample size, but it may also have caused the respondents to report positive outcomes due to feelings of appreciation for medical treatment, or as a means of thanking the clinical team for treatment. The other three studies posted the measures to the participants, which is less likely to elicit socially desirable responses (S. Harding & T. Moss, 2012a, unpublished data; S. Harding & T. Moss, 2012b, unpublished data; *Harrington, McGurk & Llewellyn, 2008*). Postal surveys can result in a low return rate, although those reviewed here received 53-55% (respectively S. Harding & T. Moss, 2013a, unpublished data; *Harrington, McGurk & Llewellyn, 2008*) and can be argued to be reasonable. A separate consideration is that they may be biased through participants self-selecting and subsequently call into the question the generalisability of the findings.

All the studies included in this review were quantitative in nature, and used previously constructed measures (Supplemental Information E). Measures such as the Medical Outcomes Short Form 12 (SF-12) have normative data that allows findings to be compared with general population (S. Harding & T. Moss, 2013a, unpublished data; S. Harding & T. Moss, 2013b, unpublished data; *Llewellyn et al., 2011*). Other measures have only been used in other disease populations, such as hospital anxiety and depression scale (*Harrington, McGurk & Llewellyn, 2008; Llewellyn et al., 2011*). An exception to this was one of the measures used in *Llewellyn et al. (2011)*. In this study, two items were derived from the EORTC QLQ-C30, which were used to assess cancer specific global Quality of Life/health status.

In medical population studies the confounding factors such as stage or exact location of tumour may be predictive factors and it is therefore important to ensure that these are appropriate selected and analysed (*Bellizzi & Blank, 2006; Brunet et al., 2010; Gallagher-Ross, 2012*). Similar factors were used across all studies included in this review

Table 3 Participants and variables.

Study	Author(s)	Participants (gender, age)	Time of measurement	Non-respondents/dropouts	Exclusion criteria	Cancer site	Cancer staging	Cancer treatments	Time since completion of treatment
1	Harrington, McGuck & Jewell (2008)	N = 76 (55% response rate; 37 Male, 39 Female; Mean Age 66.9, SD 12.6, Range 32–97; 71 White)	0–6mths = 1, 6–12mths = 3, 13–24mths = 7, 25–47mths = 20, 48–72mths = 19, and 73–121mths = 26	Significant difference between gender in responders and non-responders (more females responding)	Under 18 years of age. Having palliative treatment. Recurrent diagnosis, metastatic disease in other parts of the body (excluding neck nodes), a diagnosis of lymphoma, mental to cognitive impairments or insufficient understanding of English.	Not stated	Stage 1–2 - N = 53, Stage 3–4 - N = 23	Surgery only - N = 35, Radiotherapy only - N = 10, Surgery and Radiotherapy - N = 30, Surgery, radiotherapy and chemotherapy - N = 1	0–6mths = 1, 6–12mths = 3, 13–24mths = 7, 25–47mths = 20, 48–72mths = 19, 73–121mths = 26
2	Llewellyn et al. (2011)	T1, N = 103 (73 Males, 30 Females; Mean Age 63, SD 13.9, Range 23–91; 93 White); T2, N = 68 (Gender, Age, Ethnicity data provided)	T1 = Between diagnosis and start of treatment, T2 = 6 months after completion of treatment	There were no significant differences between patients included and not included with respect to gender, stage of cancer, 35 people did not complete the second time point. No information is given about they compared at T1.	Under 18 years of age. Having palliative treatment. Recurrent diagnosis, metastatic disease in other parts of the body (excluding neck nodes), a diagnosis of lymphoma, mental to cognitive impairments or insufficient understanding of English.	Oral Cavity - N = 68, Pharynx - N = 8, Larynx - N = 19, Other - N = 8	Stage 1 - N = 34, Stage 2 - N = 25, Stage 3 - N = 23, Stage 4 - N = 17, Missing data - N = 4	Surgery only - N = 36, Radiotherapy only - N = 25, Chemotherapy only - N = 3, Surgery and Radiotherapy - N = 17, Radiotherapy and chemotherapy - N = 13, Surgery, radiotherapy and chemotherapy - N = 9	Six months at T2
3	Ito et al. (2011)	N = 50 (21 Male, 29 Female; Mean Age 50, SD 13.06)	Mean time was 3.6yrs (SD 0.34)	No information is reported	Non-native Cantonese speakers, less than 6mths post treatment completion, recurrence	Oral Cavity, Oropharynx, gingival, floor of mouth, tongue, salivary glands, buccal mucosa, palate. Numbers at each site not stated.	Stage 1–2 - N = 41, Stage 3–4 - N = 5, Missing information - N = 4	Surgery only - N = 34, Surgery and Radiotherapy - N = 16	Mean time was 3.6yrs (SD 0.34)

4	S. Harding & T. Moss, (2013a, unpublished data)	N = 164 (55% response rate; 108 Male, 56 Female; Mean Age 67.0yrs, SD 12.5)	Mean time from completing treatment to completing questionnaires (Range 3-76; SD 21.8)	One difference was found between responders and non-responders with a greater number of people from less deprived areas returning questionnaires	Less than 3mths post treatment completion, recurrence	Oral Cavity - N = 68, Oropharynx - N = 43, Hypopharynx - N = 17, Larynx - N = 36	Stage Tis - N = 2, Stage 1 - N = 39, Stage 2 - N = 37, Stage 3 - N = 30, Stage 4 - N = 55, Missing data - N = 1	Surgery only - N = 52, Radiotherapy only - N = 35, Chemotherapy only - N = 1, Surgery and Radiotherapy - N = 35, Surgery and chemotherapy - N = 3, Radiotherapy and chemotherapy - N = 24, Surgery, radiotherapy and chemotherapy - N = 14	Mean time from completing treatment to completing questionnaires (Range 3-113; SD 27.8), TP2 = 45.1mths (Range 15-125; SD 45.1)
5	S. Harding & T. Moss, (2013b, unpublished data)	N = 163 (55% response rate; 105 Male, 58 Female; Mean Age 68.6yrs, SD 11.2)	Mean time from completing treatment to completing questionnaires (Range 3-113; SD 27.8), TP2 = 45.1mths (Range 15-125; SD 28.1)		Less than 3mths post treatment completion, recurrence	Oral Cavity - N = 75, Oropharynx - N = 33, Hypopharynx - N = 24, Larynx - N = 31	Stage Tis - N = 2, Stage 1 - N = 38, Stage 2 - N = 35, Stage 3 - N = 32, Stage 4 - N = 47, Missing data - N = 9	Surgery only - N = 48, Radiotherapy only - N = 35, Chemotherapy only - N = 0, Surgery and Radiotherapy - N = 44, Surgery and chemotherapy - N = 2, Radiotherapy and chemotherapy - N = 17, Surgery, radiotherapy and chemotherapy - N = 17	Mean time from completing treatment to completing questionnaires (Range 3-113; SD 27.8), TP2 = 45.1mths (Range 15-125; SD 45.1)

and were sourced from individual patient records and electronic hospital databases. It was therefore believed that all these would be accurate and allow for non-responder comparisons reported by Harding & Moss (2013a, unpublished data) and *Llewellyn et al. (2011)* to be authentic.

Overall the quality of the five reviewed articles are of a medium level. They represent a small total population of 343 people with HNC completing quantitative measure or sub-scales of measures. Insufficient data is presented from the combined sample size, or from anyone measure to allow for meta-analysis of the impact of treatment methodology, cancer site, or staging. Additionally the two papers by Harding and Moss (2013a, unpublished data; 2013b, unpublished data) have not undergone peer review and therefore need to be considered cautiously.

Demographic factors related to BF in HNC patients

The reviewed BF studies each collected a large number of demographic variables hypothesised as predictive or correlated with BF. *Harrington, McGurk & Llewellyn (2008)* undertook the first investigation into BF in the HNC patient population; however, they did not find any demographic variables correlating with BF. The subsequent work from the same research group (*Llewellyn et al., 2011*) found that there was a positive association between BF and being married or cohabiting and living alone, as well as with higher educational qualifications. Harding and Moss (2013a, unpublished data) added to this by finding that the younger the patient at time of diagnosis the greater the associated BF. Harding & Moss (2013b, unpublished data) longitudinal study further supported this relationship with the age at time of diagnosis being correlated with reported BF over both time periods.

Demographic factors related to PTG in HNC patients

Only one paper was identified as having specifically investigated PTG (*Ho et al., 2011*). Age and time since diagnosis did not show any significant relationship. Nor was there any significant difference in relation to religion or gender. Economic status showed significant relationship with PTG, with patients from the higher income reporting higher posttraumatic growth inventory (PTGI) scores. Education level, however, did not show any significant effect on PTG. As with BF, marital status showed significant association with PTG. Comparing married patients and patients not in a relationship showed that married patients reported higher scores on PTGI. Analysis showed that married patients reported higher total hope scores than their unmarried counterparts.

Relation of BF to disease characteristic and psychosocial factors in HNC patients

Harrington, McGurk & Llewellyn (2008) found that dispositional optimism and positive reframing could account for 23% of variance in BF and additionally that higher levels of religious coping was correlated with greater BF. *Harrington, McGurk & Llewellyn (2008)* did not find any relationship between BF and Anxiety, Depression, Time since treatment, Treatment, Stage of Cancer or diagnosis of further illness and this pattern was reinforced

by the findings of *Llewellyn et al. (2011)*. *Llewellyn et al. (2011)* supported the finding related to dispositional optimism and positive reframing, but also found that an increased use of emotional support and a decrease in self-blame positively affect BF. This combination of factors was found to account of 39% of BF variance. Harding and Moss (2013a, unpublished data) investigates subscales of BF; (1) 'Perceived changes in self' (2) 'Changes in interpersonal relationships' and (3) 'Changes in spirituality or the philosophy of life' using the Silver Lining Questionnaire (SLQ-Sp). They found that the less pain the patient is experiencing the more PTG they report across all three domains. Other significant correlations found within the SLQ showed that when participants did not suffer with movement restrictions, they reported greater changes in SLQ. Greater SLQ was experienced by people whose mood 'is excellent and unaffected by their cancer and also those who are 'as active as 'they' have ever been'.

Llewellyn et al. (2011) found that an increase in emotional growth was negatively related to the mental component summary (MCS) score. This indicates that higher levels of emotional growth are associated with poorer mental health related Quality of Life. This pattern is supported by Harding and Moss (2013a, unpublished data) who also found that MCS in HNC treated patients was significantly worse than the normative population. However, Harding & Moss (2013b, unpublished data) failed to find this pattern with the MCS longitudinally, in fact the 'mood' subscale of the University of Washington (UoW) scale suggested that the less the individuals mood is disturbed by their cancer the more BF they report. The same pattern was found with the 'activity' and 'recreation' sub scale of UoW.

Relation of PTG to disease characteristic and psychosocial factors in HNC patients

Ho et al. (2011) found that patients with more advanced cancer stages III and IV reported lower levels of PTG, but that different treatment modalities did not significantly influence PTG. Regarding the hope scale, the life orientation test-revised, and the PTGI correlation showed a positive relationship between hope and optimism. Both, hope and optimism are positively correlated to PTGI.

Results of regression analyses comparing hope and optimism in relation to PTG found that hope and optimism contributed to a 25% variance of PTG. However, only hope was a significant individual indicator of PTG.

DISCUSSION

The primary aim of this review was to evaluate the evidence which assesses the potential relationship between BF/PTG and medical, social and psychological variables as measured by validated scales people who have suffered from HNC. Posttraumatic growth is a rapidly developing field of research (*Larick & Graf, 2012; Kunst, 2012; Li et al., 2012*), but new and developing in the particular patient cohort (HNC) selected for this systematic review.

Because it has been suggested that BF and PTG are conceptually different constructs the authors looked at the BF manuscripts separately (S. Harding & T. Moss, 2013a,

unpublished data, S. Harding & T. Moss, 2013b, unpublished data, Harrington, McGurk & Llewellyn, 2008, Llewellyn et al., 2011) to the PTG manuscript (Ho et al., 2011). However, the demographic factors across the papers show a similar pattern of relationships across the constructs; that higher education/qualification and cohabitation/marriage are both associated with reported increased BF/PTG. Similarly, there is overlap with BF/PTG in HNC patients with disease characteristics and psychosocial factors where hope and optimism are both positively correlated with increased reported BF/PTG. Very few associations were observed with any of the HNC biomedical or disease factors and BF/PTG.

Methodological limitations of this paper

Although clear systematic criteria were used for search and inclusion strategies, it is possible that a number of biases may enter into the process by way of variations in definitions (e.g., of the BF and/or PTG construct), and in general by the narrow inclusion criteria. For example, by including quantitative empirical studies only, the possibility of deriving a fuller understanding of the mechanisms underlying any relationships between PTG and HNC remains limited. However, for the purposes of this review, we focused on the given inclusion criteria in order to carefully accumulate the literature on PTG and HNC with a view to developing a picture of the current status of empirical findings.

The limited number of the studies available for review makes it difficult to draw firm conclusions and develop hypotheses about how differing characteristics and conditions may lead to BF/PTG, and how they may inform future post-treatment interventions to encourage positive psychosocial outcomes. The inclusion of unpublished data is always a point for specific consideration, however, in this review the unpublished data was provided in addition to published data on BF. The unpublished data was specifically considering the phenomenon in question and was not given undue weight in analysis. For this reason it has been included, but rightly noted as a limitation.

In this review the primary author (SH) reviewed and evaluated all the retrieved abstracts and selected papers with twenty percent checks undertaken by co-authors. In addition the two manuscripts by the authors of this review (SH & TM), were reviewed by independent peer reviewers. This procedure has previously been validated by the Agency for Healthcare Research and Quality (Hartling et al., 2012).

The results are important contributions to the limited information available on both PTG and BF in HNC survivors. The overlapping patterns observed between the PTG and BF studies suggest that simultaneous study of the two concepts would provide insight into the conceptual distinction. Mols et al. (2009) point out that the impact of cancer in long-term survivors differs widely among individuals, and a significant number of them suffer from the negative effects of disease, where as others report significant positive effect. This dichotomy of concepts should be familiar to all allied health care professionals, but they should be mindful of the potential consequences of trying to impose expectations of patients (Bellizzi & Blank, 2006). In relation to developing an intervention it is important to identify patient characteristics (e.g., optimism, returning to

work, life satisfaction) that can be manipulated in order to promote BF and PTG. If these characteristics are known, theory driven interventions may be developed to alter them and reduce risk of negative effects and increase positive ones.

Limitations of reviewed studies

Results stemming from these studies are valuable; however, some limitations and methodological considerations should be noted. First, three of the five studies were cross-sectional in design, thus they provided the authors with limited knowledge about the temporal course of the conditions and the direction of causality between them and the related factors. It has been suggested by some models that it is the time of diagnosis that can be the onset stimulus (*Doka, 2008; Morse, 1997*), but no firm evidence has been forthcoming. This makes it difficult to draw conclusions from the findings of *Llewellyn et al. (2011)* because it may be that simply diagnosing cancer is significant enough to start patients BF which is sustained through to six months post treatment, therefore explaining the lack of difference found between the two time points. Additionally, it is not obvious whether time since diagnosis has an effect on the development of BF/PTG; only a longitudinal study would allow researchers to draw firmer conclusions about the role each suggested factor plays in the onset of PTG.

Moreover, because four studies were asking the patients retrospective questions, the possibility of distortion of results from recall bias is increased. It is possible that a patient cannot remember exactly how much support they received, for example, lifts to the hospital, people waiting for them during treatment, collection of medication from pharmacists, picking up shopping supplies. The reviewed studies relied on self-reported measures, which might be susceptible to reporting bias, according to the participant's mood or opinion or even as a result of post hoc bolstering (*Zoellner & Maercker, 2006*), thus possibly enhancing the likelihood of distorted results and the requirement for sufficiently large sample populations to account for the variability that this may introduce.

The measures used (Supplemental Information E), though being psychometrically validated, also have some restrictions. *Llewellyn et al. (2011)* used two items from the EORTC QLQ-C30, which leads to questionable interpretation of the data, as the items have been de-contextualised and therefore no longer actually measure what they claim. The Benefit Finding Scale incorporates both positively and negatively phrased items into questionnaires. The purpose for this is to counter the effects of social desirability and acquiescence (*Nunnally, 1978*). However statistical analysis of this scale has found that respondents answered the negatively phrased items differently to the positively phrased items, affecting score validity. *Schriesheim & Eisenbach (1995)* have subsequently identified three important assumptions underlying the use of balanced scales. First, acquiescence is a serious threat to the validity of score interpretation. Second, the negatively worded and positively worded items are bipolar statements within the same construct. Third, negatively worded items can be used without major adverse side-effects on the psychometric properties of the instrument. However, this may only become apparent when items are subjected to factor analysis in future work.

Another methodological limitation is that statistical analyses of studies searched only for linear relationships between BF/PTG and relevant variables. Some investigators have found curvilinear relationships between PTG and psychosocial variables might be present, for example between level of distress and BF (Lechner *et al.*, 2006) and mental health and well-being (Seery, 2011). An additional advance that could be made would be to use a control group of healthy participants to determine whether the positive changes reported stemmed from the trauma, or were simply the normal effect of time passing (e.g., aging), which affects individuals in multiple ways.

It is also worthwhile discussing some limitations regarding the samples examined in the included studies. The three published studies recruited (or retained for analysis) small sample sizes of fewer than 100 participants (Harrington, McGurk & Llewellyn, 2008; Ho *et al.*, 2011; Llewellyn *et al.*, 2011). It is recommended that for each variable being measured at least 10 participants be recruited (Pallant, 2010) and that a more conservative level of significance (e.g., $P \leq 0.001$ instead of $P \leq 0.05$) be required before conclusions can be drawn. The limitation with the small sample size studies is that the large number of variables being assessed may introduce Type I errors. Three of the five studies followed the sample size guidance (S. Harding & T. Moss, 2013a, unpublished data; S. Harding & T. Moss, 2013b, unpublished data; Llewellyn *et al.*, 2011). By contrast, the Harrington, McGurk & Llewellyn (2008) study may have failed to find statistically significant differences as the analysis of 76 respondents is likely to under-powered; with 15 variables the Wilson Van Voorhis & Morgan (2013) guidelines suggest a minimum of 105 respondents for correlation and 300 for factor analysis.

Another issue is that all the studies relied on convenience samples of volunteers in which minorities were under-represented, and relatively homogeneous samples were recruited, which challenges the generalisability of the findings. Additionally there were differences in relation to socio-economic status and ethnicity across people that responded and those that did not respond to the postal surveys. The lower recruitment rates of postal surveys to clinic surveys may be due to perceived pressure felt by people at clinic appointments. It is possible that these different methodologies affect how the questionnaires are completed and consequently the findings. However due to the small sample sizes and limited number of studies, no directional hypothesis can be made.

Future Directions

As CASP (Critical Appraisal Skills Programme, 2011) notes 'one observational study rarely provides sufficiently robust evidence to recommend changes to clinical practice or within health policy decision making'. The present review offers a summary of the limited work on BF and PTG research in relation to HNC treatment.

Future research might usefully focus on providing a review of qualitative studies in this area in order to generate further hypotheses reflecting the possible association between BF, PTG and HNC. Within the current review careful attempts were made to complement this method with objective criteria (e.g., using the 'Cohort' checklist from CASP for evaluation purposes), and to conduct the review in a manner most amenable to replication.

As with all empirical studies, the present review itself should be considered in light of other reviews (e.g., narrative) that also aim to synthesise the literature in similar and connected areas. It is also acknowledged that the evaluation of the final sample of papers draws an overly critical picture of the current status of research in this area. For example, it would be very difficult for any single study to have scored full marks on all sections of the evaluation criteria. Nevertheless, each of the papers reviewed represents an important contribution to BF/PTG research.

Questions regarding PTG definition have been mentioned, and clarification is a priority, prior to advancing research in understanding BF and PTG development, progression and model-building. Nine specific issues to arise from this heterogeneity of this area of study are given below: (1) the amount of time passed since trauma; (2) demographic variables such as age, gender, and socioeconomic status; (3) medical treatment variations, i.e., seven potential combinations of surgery, radiotherapy and chemotherapy; (4) potential intervening variables that may influence BF/PTG (e.g., emotional support, internal resources such as optimism and resilience); (5) possible confound of current (measured) BF/PTG with prior BF/PTG experiences in response to prior traumatic exposure; (6) the value of using a cut-off score to represent BF/PTG versus the value of a one-item endorsement to represent BF/PTG; (7) indication of illness as representing actual perceived traumatic stress; (8) measurement of BF/PTG as a multi-dimensional versus a general growth construct; and (9) transition between BF to PTG if indeed that occurs.

A number of key conceptual issues related to construct specification can be identified and have yet to be investigated in the reviewed HNC studies. These include the identification of pre- and post-trauma functioning. Determination of whether BF/PTG has occurred in the aftermath of trauma needs to be distinct from an identification of whether it was simply adaptive or superior coping (BF) or the reshaping of self (PTG) that took place. Moreover, identification of BF/PTG through self-report measures might be supplemented with interviews and/or measures for significant others (e.g., family, caregivers). This would enable triangulation of factors and allow for the identification of areas of superior functioning, whether cognitive or behavioural. Qualitative studies would be beneficial in exploring an individual's history in order to identify any previous trauma, prior coping strategies, resultant PTSD, BF, or PTG that may have occurred, in order to distinguish present psychological coping from past (but possibly ongoing) BF/PTG. An immediate possible way forward in the investigation of BF/PTG would be to conduct between-groups analysis (BF/PTG and non-BF/PTG group) in order to highlight the unique aspects of BF/PTG and the possible benefits that growth may confer. The first step in achieving this would be to assign a value to each measure over which a diagnosis of BF/PTG can be made. The development of the various domains within PTG and cut-offs, might be a focus for future investigations. An example, in health contexts and specifically within cancer, is growth more likely to occur earlier in some domains (e.g., appreciation of life) than in others (e.g., personal strength)? These are important contextual variable that may influence the factors involved in the emergence of BF/PTG in health contexts.

CONCLUSION

The five included papers showed a similar pattern of demographic relationships across both constructs of BF and PTG. Similarly, there is overlap with BF/PTG in HNC patients with disease characteristics and psychosocial factors. To enable a fuller understanding of these construct in HNC patients, longitudinal assessment is required using validated measures designed to assess BF & PTG.

ACKNOWLEDGEMENTS

The authors would like to give their thanks to Ms Chris Jarrett, Senior Assistant Librarian, University of the West of England, Mr David Courtney, Oral & Maxillofacial Surgery Consultant, Derriford Hospital, Plymouth and Dr John Bradford for assistance in undertaking this systematic review.

ADDITIONAL INFORMATION AND DECLARATIONS

Funding

This manuscript was prepared as part of the self-funded Professional Doctorate in Health Psychology of the first author. The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

Competing Interests

None of the authors have any financial, non-financial, professional or personal relationships, which may be considered a competing interest. Timothy Moss is an Academic Editor for PeerJ.

Author Contributions

- Sam Harding conceived and designed the experiments, performed the experiments, analyzed the data, wrote the paper.
- Fatimeh Sanipour quality checked the data extraction process and outcomes.
- Timothy Moss conceived and designed the experiments, wrote the paper.

Supplemental Information

Supplemental information for this article can be found online at <http://dx.doi.org/10.7717/peerj.256>.

REFERENCES

- Affleck G, Tennen H. 1996. Construing benefits from adversity: adaptational significance and dispositional underpinnings. *Journal of Personality* 64:899–922. DOI 10.1111/j.1467-6494.1996.tb00948.x.
- Alter CL, Pelcovitz D, Axelrod A, Goldenberg B, Harris H, Meyers B, Grobois B, Mandel F, Septimus A, Kaplan S. 1996. Identification of PTSD in cancer survivors. *Psychosomatics* 37:137–143 DOI 10.1016/S0033-3182(96)71580-3.
- American Psychiatric Association. 2013. *Diagnostic and statistical manual of mental disorders*. 5th ed. Washington, DC: American Psychiatric Association, 271–280. Available at www.DSM5.org.

- Andrykowski MA, Cordova MJ, Studts JL, Miller TW. 1998. Posttraumatic stress disorder after treatment for breast cancer: prevalence of diagnosis and use of the PTSD Checklist-Civilian Version (PCL-C) as a screening instrument. *Journal of Consulting and Clinical Psychology* 66:586–590 DOI 10.1037/0022-006X.66.3.586.
- Bellizzi KM, Blank TO. 2006. Predicting posttraumatic growth in breast cancer survivors. *Health Psychology* 25:47–56 DOI 10.1037/0278-6133.25.1.47.
- Booth A, Fry-Smith A. 2004. Developing a research question. In: Petticrew M, Roberts H, eds. *Systematic reviews in the social sciences*. Oxford: Blackwell.
- Brennan J. 2001. Adjustment to cancer - coping or personal transition? *Psychooncology* 10:1–18 DOI 10.1002/1099-1611(200101/02)10:1<1::AID-PON484>3.0.CO;2-T.
- Brennan J, Moynihan C. 2004. *Cancer in Context: a practical guide to supportive care*. Oxford: Open University Press.
- Brunet J, McDonough MH, Hadd V, Crocker PR, Sabiston CM. 2010. The posttraumatic growth inventory: an examination of the factor structure and invariance among breast cancer survivors. *Psycho-Oncology* 19(8):830–838 DOI 10.1002/pon.1640.
- Calhoun LG, Cann A, Tedeschi RG, McMillan J. 2000. A correlational test of the relationship between posttraumatic growth, religion, and cognitive processing. *Journal of Traumatic Stress* 13:521–527 DOI 10.1023/A:1007745627077.
- Calhoun LG, Tedeschi RG. 1998. Posttraumatic growth: future directions. In: Tedeschi RG, Park CL, Calhoun LG, eds. *Posttraumatic growth: positive changes in the aftermath of crisis*. Mahwah, NJ: Lawrence Erlbaum Associates, 215–238.
- Cancer Research UK. 2013. CancerStats: Cancer Statistics for the UK. Available at <http://www.cancerresearchuk.org/cancer-info/cancerstats/> (accessed 22 June 2013).
- Carver CS, Antoni MH. 2004. Finding benefit in breast cancer during the year after diagnosis predicts better adjustment 5 to 8 years after diagnosis. *Health Psychology* 23:595–598 DOI 10.1037/0278-6133.23.6.595.
- Collins RL, Taylor SE, Skokan LA. 1990. A better world or a shattered vision? Changes in life perspectives following victimization. *Social Cognition* 8:263–285 DOI 10.1521/soco.1990.8.3.263.
- Critical Appraisal Skills Programme. 2011. Making Sense of Evidence. Available at <http://www.casp-uk.net/>.
- Doka KJ. 2008. *Counseling individuals with life-threatening illness*. New York: Springer.
- Eysenck MW. 1992. *Anxiety: The Cognitive Perspective*. Hove, UK: Lawrence Erlbaum.
- Gallagher-Ross S. 2012. Predictors of posttraumatic growth in breast cancer survivors: An analysis of hardiness, attachment, and cognitive appraisal. *ETD Collection for Fordham University. Paper AAI3452789*.
- Harrington S, McGurk M, Llewellyn CD. 2008. Positive consequences of head and neck cancer: key correlates of finding benefit. *Journal of Psychosocial Oncology* 26:43–62 DOI 10.1080/07347330802115848.
- Hartling L, Hamm M, Milne A, Vandermeer B, Santaguida PL, Ansari M, Tsertsvadze A, Hempel S, Shekelle P, Dryden DM. 2012. *Validity and inter-rater reliability testing of quality assessment instruments (Rep. No. 12-EHC039-EF)*. Rockville, USA: Agency for Healthcare Research and Quality.
- Ho S, Rajandram RK, Chan N, Samman N, McGrath C, Zwahlen RA. 2011. The roles of hope and optimism on posttraumatic growth in oral cavity cancer patients. *Oral Oncology* 47:121–124 DOI 10.1016/j.oraloncology.2010.11.015.

- Horney DJ, Smith HE, McGurk M, Weinman J, Herold J, Altman K, Llewellyn CD. 2011. Associations between quality of life, coping styles, optimism, and anxiety and depression in pretreatment patients with head and neck cancer. *Head & Neck* 33:65–71 DOI 10.1002/hed.21407.
- Horowitz MJ. 1986. *Stress response syndrome*. New York: Jason Aronson.
- Kunst MJ. 2012. Recalled peritraumatic distress in survivors of violent crime: exploring its impact on the relationship between posttraumatic stress disorder symptoms and posttraumatic growth. *The Journal of Nervous and Mental Disease* 200:962–966 DOI 10.1097/NMD.0b013e3182718a74.
- Larick JG, Graf NM. 2012. Battlefield compassion and posttraumatic growth in combat servicepersons. *Journal of Social Work Disability Rehabilitation* 11:219–239 DOI 10.1080/1536710X.2012.730824.
- Lechner SC, Carver CS, Antoni MH, Weaver KE, Phillips KM. 2006. Curvilinear associations between benefit finding and psychosocial adjustment to breast cancer. *Journal of Consultant Clinical Psychology* 74:828–840 DOI 10.1037/0022-006X.74.5.828.
- Leventhal H, Nerenz DR, Steele DE. 1984. Illness representations and coping with health threats. In: Baum A, Singer J, eds. *A handbook of psychology and health*. Hillsdale, NJ: Erlbaum, 219–252.
- Li Y, Cao F, Cao D, Wang Q, Cui N. 2012. Predictors of posttraumatic growth among parents of children undergoing inpatient corrective surgery for congenital disease. *Journal of Pediatric Surgery* 47:2011–2021 DOI 10.1016/j.jpedsurg.2012.07.005.
- Llewellyn CD, Horney DJ, McGurk M, Weinman J, Herold J, Altman K, Smith HE. 2011. Assessing the psychological predictors of benefit finding in patients with head and neck cancer. *Psycho-Oncology* DOI 10.1002/pon.2065.
- Mols F, Vingerhoets AJ, Coebergh JW, van de Poll-Franse LV. 2009. Well-being, posttraumatic growth and benefit finding in long-term breast cancer survivors. *Psychology & Health* 24:583–595 DOI 10.1080/08870440701671362.
- Morse JM. 1997. Responding to threats to integrity of self. *Advances in Nursing Science* 19:21–36 DOI 10.1097/00012272-199706000-00003.
- Morse JM, Johnson JL. 1991. Towards a theory of illness: the illness constellation model. In: Morse JM, Johnson JL, eds. *The illness experience*. London: Sage, 315–342.
- Moyer A, Salovey P. 1996. Psychosocial sequelae of breast cancer and its treatment. *Annals of Behavioral Medicine* 18:110–125 DOI 10.1007/BF02909583.
- National Institute for Clinical Excellence. 2004. *Guideline development methods: information for national collaborating centres and guideline developers*. London: National Institute for Clinical Excellence. Available at http://www.nice.org.uk/niceMedia/pdf/GDM_Allchapters_0305.pdf.
- Nunnally JC. 1978. *Psychometric theory*. 2nd edn. New York: McGraw-Hill.
- Pallant J. 2010. *SPSS survival manual: A step by step guide to data analysis using SPSS*. 4 edn. Open University Press.
- Park CL, Ai AL. 2006. Meaning-making and growth: new direction for research on survivors of trauma. *Journal of Loss and Trauma* 11:389–407 DOI 10.1080/15325020600685295.
- Paton D. 2006. Posttraumatic growth in disaster and emergency work. In: Calhoun LG, Tedeschi RG, eds. *Handbook of posttraumatic growth: research and practice*. Lawrence Erlbaum Associates, p. 243.
- Petrie KJ, Buick DL, Weinman J, Booth RJ. 1999. Positive effects of illness reported by myocardial infarction and breast cancer patients. *Journal of Psychosomatic Research* 47:537–543 DOI 10.1016/S0022-3999(99)00054-9.

- Petticrew M, Roberts H. 2006. How to appraise the studies: an introduction to assessing study quality. In: Petticrew M, Roberts H, eds. *Systematic reviews in the social sciences: a practical guide*. Malden, MA: Blackwell Publishing, 125–163.
- Schriesheim CA, Eisenbach RJ. 1995. An exploratory and confirmatory factor-analytic investigation of item wording effects on the obtained factor structures of survey questionnaire measures. *Journal of Management* 21:1177–1193 DOI 10.1177/014920639502100609.
- Schulz R, Williamson GM, Knapp JE, Bookwala J, Lave J, Fello M. 1995. The psychological, social and economic impact of illness among patients with recurrent cancer. *Journal of Psychosocial Oncology* 13:21–45 DOI 10.1300/J077V13N0302.
- Schulz U, Mohamed NE. 2004. Turning the tide: benefit finding after cancer surgery. *Social Science and Medicine* 59:653–662 DOI 10.1016/j.socscimed.2003.11.019.
- Sears SR, Stanton AL, Danoff-Burg S. 2003. The yellow brick road and the emerald city: benefit finding, positive reappraisal coping and posttraumatic growth in women with early-stage breast cancer. *Health Psychology* 11:487–497 DOI 10.1037/0278-6133.22.5.487.
- Seery MD. 2011. Resilience: a silver lining to experiencing adverse life events? *Psychological Science* 20:390–394.
- Stanton AL, Bower JE, Low CA. 2006. Posttraumatic growth after cancer. In: Calhoun LG, Tedeschi RG, eds. *Handbook of posttraumatic growth: research and practice*. Mahwah, NJ: Erlbaum, 138–175.
- Stoll C, Schelling G, Goetz AE, Kilger E, Bayer A, Kapfhammer HP, Rothenhausler HB, Kreuzer E, Reichart B, Peter K. 2000. health-related quality of life and post-traumatic stress disorder in patients after cardiac surgery and intensive care treatment. *Journal of Thoracic and Cardiovascular Surgery* 120:505–512 DOI 10.1067/mtc.2000.108162.
- Sumalla EC, Ochoa C, Blanco I. 2009. Posttraumatic growth in cancer: reality or illusion? *Clinical Psychology Review* 29(1):24–33 DOI 10.1016/j.cpr.2008.09.006.
- Tedeschi RG, Calhoun LG. 1996. The posttraumatic growth inventory: measuring the positive legacy of trauma. *Journal of Trauma Stress* 9:455–472 DOI 10.1002/jts.2490090305.
- Tennen H, Affleck G. 2002. Benefit-finding and benefit-reminding. In: Snyder CR, Lopez SJ, eds. *The handbook of positive psychology*. New York: Oxford University Press, 584–594.
- The Cochrane Collaboration. 1999. Cochrane handbook. <http://hiru.mcmaster.ca/cochrane/cochrane/hbook.htm> [On-line].
- Thomson ResearchSoft. 2000. Reference Manager [Computer software].
- Timberlake N, Klinger L, Smith P, Venn G, Treasure T, Harrison M, Newman SP. 1997. Incidence and patterns of depression following coronary artery bypass graft surgery. *Journal of Psychosomatic Research* 43:197–207 DOI 10.1016/S0022-3999(96)00002-5.
- Tomich PL, Helgeson VS. 2004. Is finding something good in the bad always good? Benefit finding among women with breast cancer. *Health Psychology* 23:16–23 DOI 10.1037/0278-6133.23.1.16.
- Wilson Van Voorhis CR, Morgan BL. 2013. Understanding power and rules of thumb for determining sample sizes. *Tutorials in Quatitative Methods for Psychology* 3(2):43–50.
- Zoellner T, Maercker A. 2006. Posttraumatic growth in clinical psychology – A critical review and introduction of a two component model. *Clinical Psychology Review* 26:626–653 DOI 10.1016/j.cpr.2006.01.008.

FURTHER READING

- Antoni MH, Lehman JM, Kilbourn KM, Culver JL, Alferi SM, Yount SE, McGregor BA, Arena PL, Harris SD, Price AA, Carver CS. 2001. Cognitive-behavioural stress management intervention decreases the prevalence of depression and enhances benefit finding among women under treatment for early-stage breast cancer. *Health Psychology* 20(1):20–32 DOI 10.1037/0278-6133.20.1.20.
- Bostock L, Sheikh AI, Barton S. 2009. Posttraumatic growth and optimism in health-related trauma: a systematic review. *Journal of Clinical Psychology in Medical Settings* 16:281–296 DOI 10.1007/s10880-009-9175-6.
- Carver CS. 1997. You want to measure coping but your protocol's too long: consider the Brief COPE. *International Journal of Behavioural Medicine* 4:92–100 DOI 10.1207/s15327558ijbm04016.
- Hassan SJ, Weymuller EA Jr. 1993. Assessment of quality of life in head and neck cancer patients. *Head & Neck* 15:485–496 DOI 10.1002/hed.2880150603.
- Ho SM, Chan CL, Ho RT. 2004. Posttraumatic growth in Chinese cancer survivors. *Psycho-Oncology* 13:377–389 DOI 10.1002/pon.758.
- Ho SMY, Ho JWC, Bonanno GA, Chu ATW, Chan EMS. 2010. Hopefulness predicts resilience after hereditary colorectal cancer genetic testing: a prospective outcome trajectories study. *BMC Cancer* 10:279 DOI 10.1186/1471-2407-10-279.
- Lai JCL, Cheung H, Lee WM, Yu H. 1998. The utility of the revised Life Orientation Test to measure optimism among Hong Kong Chinese. *International Journal of Psychology* 33:45–56 DOI 10.1080/002075998400600.
- Melville MR, Lari MA, Brown N, Young T, Gray D. 2003. Quality of life assessment using the short form 12 questionnaire is as reliable and sensitive as the short form 36 in distinguishing symptom severity in myocardial infarction survivors. *Heart* 89:1445–1446 DOI 10.1136/heart.89.12.1445.
- Rogers SN, Gwanne S, Lowe D, Humphris G, Yueh B, Weymuller EA Jr. 2002. The addition of mood and anxiety domains to the University of Washington quality of life scale. *Head & Neck* 24:521–529 DOI 10.1002/hed.10106.
- Rogers SN, Lowe D, Brown JS, Vaughan ED. 2001. The relationship between length of stay and health-related quality of life in patients treated by primary surgery for oral and oropharyngeal cancer. *International Journal of Oral and Maxillofacial Surgery* 30:209–215 DOI 10.1054/ijom.2001.0040.
- Rogers SN, Lowe D, Humphris G. 2000. Distinct patient groups in oral cancer: a prospective study of perceived health status following primary surgery. *Oral Oncology* 36:529–538 DOI 10.1016/S1368-8375(00)00046-4.
- Scheier MF, Carver CS. 1985. Optimism, coping, and health assessment and implications of generalized outcome expectancies. *Health Psychology* 4:219–247 DOI 10.1037//0278-6133.4.3.219.
- Snyder CR, Simpson SC, Michael ST, Cheavens J. 2001. *Optimism and hope constructs: variant on a positive expectancy theme*. Washington, DC: American Psychological Association.
- Sodergren SC, Hyland ME. 2000. What are the positive consequences of illness? *Psychology and Health* 15:85–97 DOI 10.1080/08870440008400290.
- Sodergren SC, Hyland ME, Singh SJ, Sewell L. 2002. The effect of rehabilitation on positive interpretations of illness. *Psychology and Health* 17:753–760 DOI 10.1080/0887044021000009674.

-
- Ware J Jr, Kosinski M, Keller SD. 1996. A 12-item short-form health survey: construction of scales and preliminary tests of reliability and validity. *Medical Care* 34:220–233
DOI 10.1097/00005650-199603000-00003.
- Zigmond AS, Snaith RP. 1983. The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica* 67:361–370 DOI 10.1111/j.1600-0447.1983.tb09716.x.

6.1.4 Study Limitations

This was the first systematic review I had undertaken and was completed following a Master's level training course on performing these and meta-analysis studies. It was therefore interesting to receive a reviewer's comment suggesting that I wait and re-do the review when more work had been published:

I commend the authors for their effort in compiling the literature on this important topic. However, my main issue with this manuscript is that the review includes only 3 published articles and 2 unpublished manuscripts (which are written by the review authors). The authors also acknowledge this as a major limitation of the study. However, it seems to me that research on benefit finding and posttraumatic growth in people who have experienced head and neck cancer is not yet developed or ripe enough for a systematic review. Although the general methods used in the current study seem sound, with the exception for including unpublished data and only one author performing the main study screening, I recommend the authors to wait with trying to publish this manuscript in its current form until more studies have been published in this field. However, if the authors indeed want to publish the current findings I would advise them to frame their write up more as a conceptual and argumentative paper that could include an informal review of the current literature, rather than a formal systematic review.

My response was:

We share the reviewer's frustration that there is not more high quality research in this field. However, we do not believe that this detracts from the validity of conducting a systematic review. We are reminded of the original purpose of conducting and reporting a systematic review. This is to evaluate the extent of published evidence, and consider the quality of that evidence. The finding that there is little published that meets the inclusion criteria is in itself important. It is not uncommon for systematic reviews to be published with a small number of included studies, and we

are aware of at least one systematic review published with no studies meeting the inclusion criteria (Mogan and Vogel, 2009).

Of course the reviewer makes a valid point, and even after completing my research reported in paper 6, there were still only 8 papers published in this area with people who had had HNC. An issue raised by the reviewer is also that of grey or unpublished literature. The two pieces of grey literature cited in this systematic review were both by me, and as can be seen from this thesis, it took another 6 years for this work to be published.

All three systematic reviews reported in this chapter retain solely papers that use quantifiable data. While there are qualitative systematic reviews, I think it is methodologically more challenging to manage this type of data. This is increasingly complicated by the new types of data that researchers can access. Qualitative researchers may find blogs and vlogs to be valuable sources of information for investigating human health experiences. However, they present a challenge in their analysis, as they may include distinctly different types of information from the traditional methodological and validity type questions.

From a researcher perspective, using blogs/vlogs and web fora allows for 'naturalistic' data collection without interfering with the natural process of data creation, which is fully done in interaction with other web forum users rather than via a researcher (Tinati et al 2014). Therefore, there are less likely to be self-presentation biases, biases towards the researcher's agenda are avoided and what is discussed is truly of relevance to the group under investigation. Other advantages of using these web-based sources of data include decreased research costs and a wider geographic range. The extensive geographical area allows researchers the access to greater numbers of individuals with specified health problems than would be possible in a physical study with geographic constraints. Although I find these advantages to be tempting, especially when considering investigating conditions that might be classified as 'rare', for example male breast cancer, the public and private nature of blog content, needs to be considered. Researchers need to consider their research design with regard to protection of human subjects, informed consent, and bloggers' rights to privacy (British Psychological Society, 2012). Blogs/Vlogs are simultaneously private and yet quite public (Huffington Post, 2008). The private nature is reflected in the "intimate, often ferocious expression of the blogger's

passions'' (Huffington Post, 2008, p. 6), whereas the public nature is inherent in the very fact that anyone with Internet access can read those intimate expressions.

These considerations are just some of the elements of research that need to be addressed if we are going to embrace the widening sources of available data, without even considering the collection and storage, the diverse approaches to social media as data, analytical tools, or social media tools (Sloan, 2016).

A further issue in the challenge of including work in quantitative or qualitative systematic reviews, is when the peer review article is actually a video journal such as the Journal of Visualized Experiments (e.g. <https://www.jove.com/video/54788/behavioral-assessment-hearing-2-to-4-year-old-children-two-interval>), and developing methodologically sound approaches for how these are evaluated and compared to the more traditional forms of presentation.

6.1.5 Contribution to knowledge and autoethnographic issues

6.1.5.1 Contribution to knowledge

When I undertook the review included as paper 7, the primary aim was to establish what measures were used within the field of PPC. It was also important to understand what at the time, counted as knowledge in that area (i.e., what factors were investigated and recorded in the disease group). The aim, as cited within the paper, was to establish what was currently known; the specific purpose, to argue that the subsequent research was relatable to previous research, but also contributing to knowledge. I believe that this was achieved, and allowed me to relate my own work to the literature on HNC, and also understand it in wider, primarily breast cancer studies.

Appendix 17 has an overview of how the research undertaken in paper 7 was disseminated prior to the paper's publication. It also provides a list of papers where paper 7 has been cited, and this is followed by a quality appraisal of the paper.

6.1.5.2 Autoethnographic issues

This was my first systematic review. I led it from conception to publication. An element of this that I enjoyed was its process-driven nature. The precise step-by-step requirements of undertaking the review allows for multiple small 'wins'. This is unusual in the life of a researcher. We spend a lot of time working on grant writing, putting together ethics applications, undertaking analysis and writing journal articles

etc. All of these can take weeks or months to complete and may lead to nothing. The review process, however, although not always leading to a publication, provides a series of end points (run the search, de-duplicate the database, remove references at title level etc.).

During the life-cycle of this systematic review, I began working on two other reviews and these are represented by papers 8 and 9 in this thesis. These publications have led to me getting the reputation of being a go-to person for help and guidance when undertaking a systematic review. Consequently, the research and innovation department of the NHS trust I work for, advise people who ask them for help in grant development where a review is likely to be included, or when they want to undertake a review prior to a grant application, to work with me. It is lovely being identified as a person with a specific skill set, but I am conscious of not wanting to become pigeon holed.

6.1.6 Future research questions

The questions at the end of the systematic review reported in paper 7, were those that led to papers 2 and 6:

- After the completion of treatment for Head and Neck Cancer, what are the demographic, clinical, and psychological factors associated with PPC that occur in the acute period, defined as between 3 and 12 months?

and

- After the completion of treatment for Head and Neck Cancer, what are the trajectories for positive psychological change longitudinally (defined as longer than 12 months)?

Specifically it was evident that people did not understand what the pattern of PPC development was in the long term.

6.2 Paper 8 - Clinical experience leading to the research work and production of the paper

I started working at the Bristol Speech and Language Therapy Research Unit in July of 2011. I was employed as a senior research assistant on an NIHR programme grant (Child Talk - RP-PG-0109-10073) which aimed to investigate speech and language therapy practices used with preschool children with speech language or communication needs, in isolation from any other disorder such as autism, or cerebral palsy. It was in this role that I met and started to work with Anna Blackwell the first author of paper 8. Anna was funded as part of Child Talk to undertake a PhD, which looked at the evolving language environments of preschool children.

Paper 8 examines the available literature using observations of parent child interaction with children with primary language impairment and their typically developing peers. The extent of parent child interaction differences between these groups has implications for the use of parent child interaction interventions and for research into the relationship between children's environment and their language development.

Anna and I worked together to develop the search strategy and strings. We then worked through the review stages together. This included reaching consensus about inclusion of papers and quality appraisal scores. It made sense that I was the second reviewer as the systematic review covered work Anna needed for her thesis.

6.2.1 Research Questions

The aim of this review was to identify whether or not there are differences in the characteristics of parent child interaction between preschool children with language delay and their typically developing peers.

6.2.2 Study Design

The same study design presented in Section 6.1.2 was completed for paper 8. The key difference being the selection of the search terms was conducted in collaboration with Anna Blackwell.

6.2.3 Characteristics of Parent-Child Interactions: A systematic review of studies comparing children with primary language impairment and their typically developing peers

Characteristics of Parent–Child Interactions: A Systematic Review of Studies Comparing Children With Primary Language Impairment and Their Typically Developing Peers

Communication Disorders Quarterly
1–12
© Hammill Institute on Disabilities 2014
Reprints and permissions:
sagepub.com/journalsPermissions.nav
DOI: 10.1177/1525740114540202
cdq.sagepub.com
SAGE

Anna K. M. Blackwell, BSc^{1,2}, Sam Harding, MPhil^{1,2}, Selma Babayiğit, PhD¹, and Sue Roulstone, PhD^{1,2}

Abstract

The importance of parent–child interaction (PCI) for language development has been well established. This has led many speech and language therapy (SLT) interventions to focus on modifying PCI as a means to improving children's early language delay. However, the success of such programs is mixed. The current review compares PCI, observed in naturally occurring contexts, with preschool children with language delay and age- or language-matched typically developing (TD) controls. A systematic review of the literature searched 10 databases for studies using a case-control design and extracted data concerning participants, matching, selection, design, assessments, measures, findings, statistics, and bias. Quality appraisal used the Critical Appraisal Skills Programme case-control checklist. The search identified 17,824 articles, which were reviewed against exclusion criteria. The final review included 9 studies, which were diverse in terms of matching, delay criteria, and PCI measure. A narrative synthesis was conducted. The evidence for PCI differences between children with language delay and TD peers was limited and any suggestion that parents were less responsive could be attributed to limited language skills of children with language delay. The findings question the assumption that communicative environments of children with language delay are different, although the evidence is from a small sample of children from middle-class families. Children with language delay may instead be less able to learn from their environment. The review highlights the gap in understanding the relationship between parent and child language use during PCI. The need for further, longitudinal research is emphasized, including children ranging in type and severity of delay, across diverse socioeconomic backgrounds.

Keywords

communication, acquisition/development, language/linguistics, delays/disorders

Background

Approaches to speech and language therapy (SLT) interventions can be divided into “child-focused” and “environmental” methods; the latter are based on working with the people who interact with the child (Pickstone, Goldbart, Marshall, Rees, & Roulstone, 2009). Environmental approaches include interventions that aim to modify parent–child interaction (PCI), based on the assumption that changing the behavior of parents who interact with children can produce improvements in their language (Baxendale & Hesketh, 2003; Gibbard, 1994; Girolametto, Pearce, & Weitzman, 1996). A systematic review of the effectiveness of SLT interventions found that including parents in interventions could have beneficial effects (Law, Garrett, & Nye, 2003). However, the review found that parental response to PCI interventions was varied. For example, Fey,

Cleave, and Long (1997) found that following training, parents' use of recasting could be categorized according to the frequency with which the parents subsequently used recasts. Using more recasts was related to greater language gains for their children. Interventions that involve parent training may be more appropriate for certain families (Gibbard, 1994). An individualized approach would ensure that families enrolled in PCI interventions are those best suited to this type of program.

¹The University of the West of England (UWE), Bristol, UK

²Bristol Speech and Language Therapy Research Unit, Frenchay Hospital, Bristol, UK

Corresponding Author:

Anna K. M. Blackwell, Bristol Speech and Language Therapy Research Unit, Frenchay Hospital, Bristol, BS16 1LE, UK.
Email: Anna.Blackwell@uwe.ac.uk

Approaches that modify PCI are derived from factors found to be positively related to typically developing (TD) language (Pickstone et al., 2009). There is an abundance of research investigating features of parent language that influence the language development of TD children. Research has found striking differences in children's vocabulary skills related to parental language input: the number, diversity, and sophistication of words parents direct at their children (Huttenlocher, Haight, Bryk, Seltzer, & Lyons, 1991; Rowe, 2012). Differences in children's vocabulary sizes have also been found between high and low socioeconomic status (SES) groups, which appear to be mediated by the characteristics of parent speech (Hart & Risley, 1995; Hoff, 2003). In a longitudinal study of language development, the effects of adult language input on children's development was partially mediated by adult-child conversations, which were found to be significantly associated with language development (Zimmerman et al., 2009). These findings highlight the value of engaging children in conversation, talking directly to them and responding to their interests, beyond providing rich linguistic input (Zimmerman et al., 2009).

However, PCI strategies that support TD language skills may not be sufficient for children with delayed language development. Research has suggested that the language input that children with delayed language receive, as well as the interactions that they partake in, is different to that of TD peers, which may impact on their language development (Whitehurst, Novak, & Zorn, 1972). However, the relationship between PCI and child language development has been recognized as reciprocal (Gibbard, 1994). There is a need for a better understanding of parent and child characteristics that are related to delayed language development that can inform SLT interventions.

The current systematic review therefore focused on research that aims to identify differences in PCI with TD and language-delayed children. The review concentrated on studies with preschool children as improved PCI has been shown to be an important outcome target of therapy with children aged 2 to 3 years for speech and language therapists (Roulstone, Wren, Bakopoulou, Goodlad, & Lindsay, 2012). The review is focused on children who have an isolated difficulty with the acquisition of language, despite otherwise typical development. There is a range of terminology used by researchers and clinicians to describe these children, including "language-delayed" (Cunningham, Siegel, van der Spuy, Clark, & Row, 1985), "specific language impairment" (SLI; Fey, Krulik, Loeb, & Proctor-Williams, 1999), "slow expressive language development" (Paul & Elwood, 1991), and "late-talking" (Rescorla & Fechnay, 1996).

The diversity of terms suggests a heterogeneous condition, without commonly recognized criteria or definition (Law, Boyle, Harris, Harkness, & Nye, 2000). The choice

of term may be partly mediated by the age of the children and whether or not they have received a formal diagnosis. Children identified as having delayed language development may have only transient language difficulties and will not necessarily receive a later diagnosis of language impairment. Around half of children with language delay have been shown to "catch up" to their TD peers by 4 years old (Dale, Price, Bishop, & Plomin, 2003). However, it may not be possible to separate delayed or impaired children into two clear groups, even though this approach might be preferable because it could help interpret the results of intervention studies. Many studies use the same language inclusion criteria within a wide age range of 12 months or more (e.g., Baxendale & Hesketh, 2003; Conti-Ramsden, 1990; Paul & Elwood, 1991; Proctor-Williams, Fey, & Loeb, 2001). In these samples, the older children may be more clearly recognizable as language impaired (Paul & Elwood, 1991), or demonstrate more severe impairment than the younger children in the group. The term *primary language impairment* (PLI) was used for this review to include all of these descriptions and refers to children identified through diagnosis or study assessment as having a difficulty or delay with language, where there is no overt diagnosis of general developmental delay or, sensory or neurological disorder. Prevalence of PLI is around 6% (Law et al., 2000) and is associated with poor literacy skills and later academic, social, and behavioral problems (Beitchman et al., 2008; Conti-Ramsden, Mok, Pickles, & Durkin, 2013; Snowling, Bishop, Stothard, Chipchase, & Kaplan, 2006; St Clair, Pickles, Durkin, & Conti-Ramsden, 2011; Stothard, Snowling, Bishop, Chipchase, & Kaplan, 1998).

The current review aimed to identify whether there were differences in the characteristics of PCI with preschool children with PLI compared with their TD peers, in studies that used naturalistic observations of PCI. The extent of PCI differences between these groups has implications for the use of PCI interventions and for research into the relationship between children's communicative environment and their language development.

Method

The systematic review was guided by the principles outlined in the Cochrane Collaboration methodology (Higgins & Green, 2011), as far as they could be applied to case-control studies.

Criteria for Including Studies

Population. Preschool children aged 0;0-5;11 (years; months) only. Studies were required to include a group of children with TD language and a group with PLI, with no other suspected disorders, for example, autism or hearing impairment, and age appropriate nonverbal/developmental

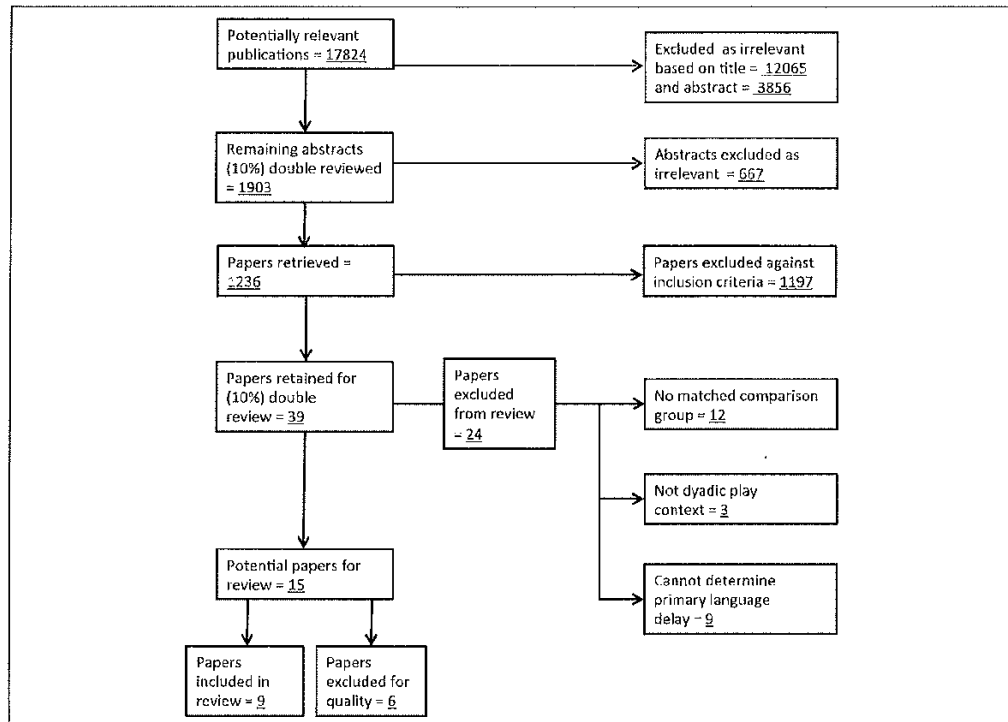


Figure 1. Flowchart of review process.

skills. Children had to be from monolingual English-speaking homes, with no reported parent mental health problems or child maltreatment.

Variable measured. Observations of dyadic PCI during play. Studies had to examine interactional characteristics of communication rather than acoustic properties of speech.

Type of study. Case-control studies only were included in the review. This decision was made to ensure at least within-study group comparisons were possible, as different characteristics of PCI were measured across studies. A separate systematic review investigating the effectiveness of interventions regarding PCI is in process as part of the Child Talk program (<http://www.speech-therapy.org.uk/projects/child-talk>); therefore, this review did not consider intervention studies.

Search Methods for Identification of Studies

Ten electronic databases were identified from their use in other systematic reviews in the field and searched (April

2012) with no date limits: MEDLINE; EMBASE; CINAHL Plus; PsycINFO; SocINDEX; PsycARTICLES; Cochrane Database of Systematic Reviews; CENTRAL; Cochrane Methodology Register; ERIC. The MEDLINE search strategy (the appendix) comprised subject headings and text words, which described the elements of the population and variable measured (outlined above) as well as exclusionary criteria. The strategy was reviewed by expert academics in the fields of language development and SLT and adapted for each database. Electronic searches were supplemented by checking references of relevant publications and included journal articles, book chapters, and doctoral dissertations (≤ 5 years old). Articles published in languages other than English were excluded due to time and resource constraints ($n = 89$).

Data Collection

The first author excluded irrelevant articles by screening titles and abstracts (see Figure 1). The remaining abstracts were fully reviewed by the first author and 10% independently checked by the second author against inclusion criteria. Any

Table 1. Methodological Quality Assessment Using CASP.

Author (year)	3 case recruitment acceptable?	4 controls acceptable?	5 Variables measured accurately?	6b. Confounders considered?	9 Results believable?	10 Can results be applied?	Quality
Conti-Ramsden and Friel-Patti (1983)	Y/CT	Y/CT	Y	Y/CT	Y/CT	Y/CT	Medium
Conti-Ramsden and Friel-Patti (1984)	As above	As above	Y	As above	Y/CT	As above	Medium
Conti-Ramsden (1990)	As above	As above	Y	As above	Y/CT	As above	Medium
Cunningham, Siegel, van der Spuy, Clark, and Bow (1985)	Y/CT	Y/CT	Y	Y	Y/CT	Y/CT	Medium
Fey, Krulik, Loeb, and Proctor-Williams (1999)	Y/CT	Y	Y	Y	Y	Y/CT	High
Proctor-Williams, Fey, and Loeb (2001)	As above	As above	Y	As above	Y	As above	High
Paul and Elwood (1991)	Y/CT	Y	Y	Y	Y	CT	Medium
Rescorla and Fechnay (1996)	CT	Y	Y	Y	Y	CT	Medium
Rescorla, Bascome, Lampard, and Feeny (2001)	CT	Y	Y	As above	Y	As above	Medium

Note. CASP = Critical Appraisal Skills Programme; Y = yes; CT = cannot tell.

disagreements were resolved through discussion and in any case of doubt the article was included in the next stage. Full text articles were then retrieved and further considered against inclusion criteria by the first author. The full text articles that were retained had relevant data extracted by the first author, using a standardized form which recorded details on participant groups and matching criteria, selection, study design, assessment tools, variables measured, main findings, statistics, and sources of bias. Questions were developed with reference to Tager-Flusberg's (2005) article on designing studies with language-disordered populations and related methodological issues. Articles were also subjected to quality assessment by the first author using the Critical Appraisal Skills Programme (CASP; 2013) case-control checklist to determine study quality, reliability, and application of findings. Studies were rated low, medium, or high quality according to the answers to CASP questions (see Table 1: Y = yes; CT = cannot tell; N = no). Quality appraisal identified six low-, seven medium-, and two high-score studies. The low-quality articles were excluded. The process was 10% independently checked by the third and fourth authors; any disagreements were discussed to establish consensus on issues of data extraction and quality appraisal.

Included studies were mixed in terms of how PLI and TD groups were matched (four chronological age and five language stage); the method for determining PLI status (clinically referred or determined by study assessment, with various criteria); the severity of children's delay; and the PCI characteristics of interest. Heterogeneity precluded meta-analysis; therefore, a narrative synthesis was used which summarized findings descriptively. To maximize the clarity of the review, Gough's (2007) "mapping stage" was implemented, by which the review area was first viewed as a whole and then in subsections. Grouping the findings

according to PCI characteristics and matching helped to guide the synthesis.

Results

Results of the Search

After removing duplicates, 17,824 articles were identified (see Figure 1). Almost 90% were excluded as irrelevant by title and abstract. The remaining abstracts ($n = 1,903$) were reviewed against inclusion criteria. For the 10% reviewed by the second author, there was agreement about the inclusion of 92% of these references. Full text articles were retrieved ($n = 1,236$) for more detailed review against inclusion criteria. Further articles were excluded because they did not include preschool children only ($n = 5$), did not assess interactional characteristics of PCI ($n = 87$), did not include the clinical population of interest ($n = 648$), or met other exclusion criteria, for example, studies of parent mental health, child maltreatment, or bilingual language learners ($n = 457$). Thirty-nine articles remained. Those without appropriately matched comparison groups ($n = 12$), observations of PCI that were not in a dyadic play context ($n = 3$), or those without clearly determined PLI ($n = 9$) were also excluded, which resulted in 15 articles. These articles used a case-controlled observational design to analyze differences in dyadic PCI, in semistructured or unstructured play settings, with preschool children with PLI and matched TD controls. Following the quality appraisal, 6 were excluded on the grounds of low methodological quality.

Included Studies

Nine studies were retained for inclusion in this review (see Table 2). Most studies used cross-sectional case-control

Table 2. Characteristics of Included Studies.

Author	Child participants	Setting	PCI variables	Findings
Conti-Ramsden and Friel-Patti (1983)	28: 14 PLI and 14 language-matched TD	15-min play videotaped in a specially designed playroom.	Meaning illocutions Cohesion illocutions Dialogue participation	Group differences: Children with PLI initiate less turns. Mothers in PLI dyads used less responsive utterances. No difference: No. of conversational turns; type of meaning illocutions used by mothers.
Conti-Ramsden and Friel-Patti (1984)	As above	As above	Dialogue analysis—initiating role and responding role	Group differences: Interaction between initiations and child language status. More topics in TD dyads. No difference: No. of conversational turns. Children all introduced more topics than mothers. All mothers initiate more than children and no difference in form or complexity.
Conti-Ramsden (1990)	As above	As above	Dialogue Participation Mothers' Contingency Coding Scheme Mothers' Speech Acts Coding System	Group differences: Mothers in PLI dyads initiated more, used less complex recasts and less cohesion illocutions when replying to their children with simple recasts or with continuations. No difference: No. of turns.
Cunningham, Siegel, van der Spuy, Clark, and Bow (1985)	60: 33 PLI and 27 age-matched TD	15-min free play and structured play videotaped in playroom.	1: Mothers responses, informal play, conversational interactions, control and reward and child compliance 2: Total no. of utterances, language complexity	Group differences: Children with PLI less likely to initiate following maternal noninteraction, increased with lower receptive scores. Mothers adjust language complexity to child's comprehension not production. Discrepancy in complexity for dyads increased with greater delay No difference: Responsiveness of mothers.
Fey, Krulik, Loeb, and Proctor-Williams (1999)	20: 10 PLI and 10 language-matched TD	2 × 30-min play videotaped eight months apart, at homes.	Reformulations Recasts: Simple or complex	No difference: Parents use of simple, complex or total recasts and recast use was stable over time.
Proctor-Williams, Fey, and Loeb (2001)	As above	3 × 30-min play videotaped four months apart, at homes.	Parental recasts - target-specific copula and/or article recasts.	Group differences: Relationship between parent copula recasts (not articles) and child copula production in TD dyads only. Children with PLI produced fewer copulas than peers. No difference: Rate of recasts.
Paul and Elwood (1991)	56: 28 PLI and 28 age-matched TD	10-min free play videotaped in designed playroom.	Mother's utterances: Syntax, pragmatic function, topic management and lexical contingency	Group differences: Fewer utterances by children and greater mother-child MLU discrepancy in PLI dyads. Mothers in TD dyads provided more expansions/ extensions. No difference: Proportion of child utterances that received expansion/ extension
Rescorla and Fechnay (1996)	36: 18 PLI and 18 age-matched TD	10-min free play videotaped with mother.	Utterance type, child compliance and communicative gestures. Coded for social cues and synchrony.	Group differences: PLI dyads showed stronger patterns of relationships between variables, for example, mother's control negatively related to synchrony and child compliance. No difference: Mother's synchrony children very similar (fewer clear verbal cues but they as "communicative").
Rescorla, Bascome, Lampard, and Feeny (2001)	53: 32 PLI and 21 age-matched TD outcomes at 36 months	10-min free play videotaped with mother.	Topic focus—synchronous and asynchronous codes Utterance function	Group differences: Mothers in PLI dyads talked more, and asked more questions. Children with PLI asked fewer questions. No difference: Children with PLI spoke as often as TD (despite lower MLU). No difference in child topic initiations.

Note. PCI = parent-child interaction; TD = typically developing; PLI = primary language impairment; MLU = mean length of utterance.

designs although two collected data longitudinally. All studies included in the final review were carried out in English-speaking countries, using English-speaking samples. Seven

articles presented analyses of the same full or partial data sets that were used in other included studies (Conti-Ramsden, 1990; Conti-Ramsden & Friel-Patti, 1983, 1984; Fey et al.,

1999; Proctor-Williams et al., 2001; Rescorla, Bascom, Lanpard, & Feeny, 2001; Rescorla & Feclnay, 1996). Overall, the review findings are based on five completely separate samples with a collective size of fewer than 150 children with PLI, and fewer than 250 children in total. Studies were published between 1983 and 2001. No studies meeting the inclusion and quality criteria were identified after 2001.

Risk of Bias

The potential for bias in the studies was related to limited details of the characteristics of the child participants and selection processes as well as parent involvement in SLT. The groups often varied in the severity of their language delay or had only mild delay. There was also concern for how accurately TD children had been matched on necessary variables.

Selecting the most appropriate comparison groups for preschool children is difficult because of their rapid development during this stage. When using TD age-matched comparisons, it is important to bear in mind that the language skills of children with PLI will be considerably below their age-matched peers. Differences in PCI between groups may therefore not be surprising and any differences could be attributed to parents adjusting to their child's language level. However, when using TD language-matched groups there is an issue about comparing more developmentally advanced children with PLI with their younger language-matched peers. No studies included in the review used both age- and language-matched comparison groups. Conclusions about study findings are therefore dependent on whether comparison groups are age- or language-matched and the synthesis of findings was grouped accordingly.

Five of the nine articles stated that children with PLI had received SLT but that these interventions did not focus on parent training (Conti-Ramsden, 1990; Conti-Ramsden & Friel-Patti, 1983, 1984; Fey et al., 1999; Proctor-Williams et al., 2001). In the remaining four articles, it was not possible to determine whether children in the PLI groups had received SLT. If parents had received training then this could exaggerate or reduce PCI differences between groups. Furthermore, parents receiving SLT sessions could have changed their interaction techniques simply from observing or discussing SLT sessions. Fey et al. (1999) raised this possibility but considered it unlikely. Parents in their study had been keen observers of SLT and yet their style of interaction was reportedly stable over time. Nonetheless, it could not be ruled out that these groups had external influences on their language behavior.

Characteristics of PCI

Broadly, the measures of PCI fell into five main categories:

1. *Quantity of language*, for example, number and rate of verbal/nonverbal acts

2. *Complexity of language*, for example, mean length of utterance (MLU)
3. *Dialogue participation*—Proportion of conversational turns and initiations
4. *Purpose of communicative act*, for example, share meaning, demonstrate intentions, maintain conversation
5. *Responsiveness*—Type and appropriateness of conversational reply in relation to previous turn, for example, elaboration and recasts.

Quantity and complexity of language. Findings regarding quantity and complexity of language came from the four studies that used age-matched controls. These studies demonstrated some differences in the amount of talk used by mothers and their children with PLI. For example, Rescorla et al. (2001) found that mothers in PLI dyads talked more than controls, while there was no group difference in the amount children communicated, in terms of total utterances, despite children with PLI having a shorter MLU. However, Paul and Elwood (1991) found that children with PLI produced fewer utterances than age-matched controls. A greater discrepancy between mother and child language complexity (MLU) was found in PLI dyads compared with control dyads (Paul & Elwood, 1991). Group differences in the language use of children with PLI are not necessarily surprising as they were recruited precisely due to lower language abilities than age expectations. Cunningham et al. (1985) found that this discrepancy in language complexity between mother and child increased with greater delay and as children interacted less. They also found that mothers in PLI dyads adjusted their language complexity to children's receptive (comprehension), rather than expressive (production) skills, which they suggested might result in parent language models that are too advanced for children to imitate.

Dialogue participation. Two studies using age-matched controls analyzed participation. They provided some evidence for group differences in child initiations. Cunningham et al. (1985) found that children with PLI initiated less following maternal noninteraction and they were more likely to ignore mothers. The study also found that younger children with PLI engaged less in interaction compared with older children with PLI and TD peers. Interaction frequency was also negatively correlated with receptive delay, as were children's initiations and responsiveness. Topic initiations, however, were found to be similar for children in both groups, with children introducing more topics than mothers (Rescorla et al., 2001).

Three language-matched control studies analyzed participation. They found no group difference in the number of conversational turns in dyads. However, children with PLI initiated less conversation than peers (Conti-Ramsden & Friel-Patti, 1983, 1984) while mothers initiated more in PLI dyads compared with controls (Conti-Ramsden, 1990). There were no differences in the form or complexity of

mother initiations between groups (Conti-Ramsden & Friel-Patti, 1984). Overall, there was a greater discrepancy in participation between partners in PLI dyads compared with control dyads. Although generally more topics were introduced in TD dyads, children in both groups were again found in these studies to introduce more topics than mothers (Conti-Ramsden & Friel-Patti, 1984).

Purpose and responsiveness of communicative acts. There were various group differences found among the four age-matched studies. Rescorla et al. (2001) found that parents of children with PLI used more questions, while their children asked less than controls. Stronger patterns of relationships were also found between variables in PLI dyads. For example, mothers' control was negatively related to synchrony and child compliance. However, there was evidence among these studies for no group differences in maternal responsiveness or synchrony (Cunningham et al., 1985; Rescorla & Fechnay, 1996). Children were also very similar across groups and although children with PLI used fewer clear verbal cues, they were as communicative as controls. Paul and Elwood (1991) highlighted the need for caution when interpreting group differences in parental responsiveness. Their study demonstrated that apparent differences in parents' expansion and extension use were no longer significant when measures were examined in relation to the proportion of child utterances.

There were some discrepancies among the five language-matched studies regarding group differences in purpose and responsiveness of utterances in dyadic interactions. Conti-Ramsden and Friel-Patti (1984) found that mothers most often responded adequately to their children across groups and all children were also found to most often respond adequately (i.e., provide clear appropriate responses when required). However, when reacting to comments, which do not require a response, children with PLI were found to be more ambiguous than peers. Mothers in PLI dyads were found to use some responsive utterances less often than mothers of TD children (Conti-Ramsden & Friel-Patti, 1983). Further analysis of this data set found a group difference in maternal contingent replies but only for complex recasts, which were used less frequently in PLI dyads (Conti-Ramsden, 1990). While there were no group differences in the use of simple recasts, when they were used, PLI group mothers used more meaning illocutions (sharing information) and less cohesion illocutions (maintaining conversational flow). There is some contention here as other studies attempting to replicate these findings demonstrated evidence for no differences in simple or complex recasts, over an 8-month period (Fey et al., 1999). Additional analysis of this 8-month data set demonstrated a relationship between parent copula recasts (*am, is, are, was*) and child copula production in TD, but not PLI dyads (Proctor-Williams et al., 2001).

Discussion

Summary of Main Findings

Heterogeneity of findings prevented clear conclusions from being drawn regarding specific PCI differences between PLI and TD dyads. However, there were some emerging trends. In particular, the findings suggested difference in dialogue participation. Children with PLI were found to initiate fewer conversational turns than their TD peers in interaction with parents. Parents in PLI dyads may consequently appear more controlling. However, children in both groups were found to introduce more topics than parents suggesting that they are allowed to guide the content. Generally, parental differences during PCI were suggested to reflect parents adjusting to the children's communicative abilities (Conti-Ramsden & Friel-Patti, 1983, 1984; Paul & Elwood, 1991), although other developmental factors such as attention (Conti-Ramsden & Friel-Patti, 1984) and behavior could also play a role.

The evidence for group differences in responsiveness was mixed. There was some evidence for group differences in recast use and the possibility that joint focus may be less common in PLI dyads (Conti-Ramsden, 1990). One study highlighted that differences in parents' responsive utterances between groups were proportional to the opportunities available to respond to the child, which were often reduced in PLI dyads (Paul & Elwood, 1991). PCI may play a role in maintaining delay. However, group differences in PCI were generally considered to be child driven. Differences in children's communicative ability may lead to the use of conversational strategies to maintain conversation (Rescorla et al., 2001). The evidence highlights the reciprocal nature of the relationship between parent and child language use. Other studies found evidence for no difference between the PLI and TD dyads. They proposed instead that the linguistic input that children with PLI receive is no less facilitative, at least in terms of recasts, but they make less efficient use of it than TD children (Fey et al., 1999; Proctor-Williams et al., 2001).

Quality of the Evidence

The systematic review highlighted a number of issues, which question the appropriateness and strength of the methodology of the included studies. Furthermore, the review did not identify recent research from the last decade that fit the inclusion and quality criteria.

Child language measures. One problem in the study of children with PLI is the appropriate definition and assessment of this population. In general, studies all sampled children with expressive language delay, which was most often measured by MLU while the use of standardized assessments varied. Both within and between studies, the children were heterogeneous in terms of their language abilities that

complicated the comparison of findings between groups as well as studies. The severity of language difficulties ranged from around 6 months to more than 2 years delay. Some of the studies that found evidence for limited or no group differences had among the most lenient inclusion criteria (Fey et al., 1999; Proctor-Williams et al., 2001; Rescorla et al., 2001; Rescorla & Fechnay, 1996). It is possible that these studies included children who had language skills better described as at the lower end of the TD spectrum, or had delayed language development but were not language impaired. This possibility was supported by the fact that some of the children in the longitudinal study later "caught up," who may be better described as "late talkers" (Fey et al., 1999), which highlights the variation in children's developmental trajectories in the early years. It is important not to use null findings to negate potentially important PCI differences for children with more severe delay or language impairment.

It is necessary to ascertain whether the children in the included studies had receptive language delay in addition to their expressive language delay. Persistence rates for children with expressive and receptive delay have been shown to be almost twice (75.6%) that of expressive-only delay (40%; Law et al., 2000). There is also less evidence that children with receptive language difficulties will respond positively to SLT interventions (Law et al., 2003). Five studies in the review stated that children's receptive language was normal, although it was not always formally assessed. Only one study clearly included children with receptive delay, which examined the influences of delay severity and found that children with more severe receptive delay were less interactive (Cunningham et al., 1985). However, three studies (Fey et al., 1999; Paul & Elwood, 1991; Proctor-Williams et al., 2001) did not mention children's receptive language ability. The lack of detail regarding children's receptive language makes it difficult to determine the extent to which PCI may be different for children with receptive language delay.

Matched comparison groups. There is a common problem in child language research regarding how best to match control groups on the variables of interest and confounders (Tager-Flusberg, 2005). The present review only considered variables to be adequately matched across groups if relevant assessment scores were provided as evidence. Accordingly, two articles (Paul & Elwood, 1991; Rescorla et al., 2001) provided sufficient evidence that groups were matched on all four variables considered: matching variable (language or age), SES, gender, and nonverbal ability. Two articles (Rescorla et al., 2001; Rescorla & Fechnay, 1996) outlined alpha levels used ($p < .05$ or $p < .001$), while the remaining articles did not mention statistical differences. Many studies assume that if assessment scores are not significantly different between groups, then variables can be

considered to be the same for each group (fail to reject null hypothesis). However, there is concern for Type II errors (fail to reject null hypothesis when in fact groups do differ). Mervis and Klein-Tasman (2004) have consequently proposed that much higher alpha levels ($p > .5$ vs. standard $p > .05$ for nonsignificance) are used for adequate matching. Exact alpha levels for nonsignificant language differences between groups were given for one data set ($p = .62$ and $.52$ at each time point; Fey et al., 1999), which suggested appropriate matching for MLU.

It is important to recognize that language is a multidimensional skill. Plante and Swisher (1993) warned that matching language on only one or a few measures, such as MLU, may undermine construct validity. Matching groups on external factors, such as SES, is also important. All articles used predominantly middle-class samples. There is a dearth of research with lower SES samples, yet these children may be at greater risk of delayed language development (Locke, Ginsborg, & Peers, 2002). Research with TD populations has highlighted a gap in children's vocabularies between higher and lower SES groups, which has been linked to less parent speech in lower SES families (Hart & Risley, 1995).

Study design. A criterion for inclusion in this review was that studies used case-control designs, which were pertinent to compare groups on a variable (PCI) that is naturally occurring. However, case-control designs can be problematic. First, there can be difficulties selecting appropriate control groups; age-matched TD controls would be expected to have greater verbal abilities, while language-matched controls would be expected to have less advanced nonverbal skills compared with PLI cases. No studies included in the review used both age- and language-matched controls, which is of critical importance as this approach could have helped to clarify whether any differences were related to children's age or language level. Second, children's TD or PLI group status precedes their involvement in the studies, most of which were cross-sectional, measuring variables at the same time point. It is therefore difficult to conclusively determine the direction of the relationship between parent and child language. According to the National Institute for Clinical Excellence (2004, updated 2005) guidelines, case-control studies "with a high risk of confounding bias, or chance and a significant risk that the relationship is not causal" should not be considered for making recommendations. Although these guidelines are for medical research, they highlight design limitations. While included studies were considered high or medium quality it should be noted that this is only within the confines of their design. The relevant issues outlined caution the evaluation of these findings as robust evidence for the existence, or lack, of PCI differences between groups or the direction of influence between parent and child.

Table 3. Retrospective Statistical Calculations.

Author	Variable (test, alpha level)	PLI <i>M</i> (SD)	Comparison <i>M</i> (SD)	Cohen's <i>d</i>	Power (group sample size)
Conti-Ramsden and Friel-Patti (1983)	Cohesion illocutions:	0.29 (0.61)	1.57 (1.74)	0.98	0.47 (14)
	Choice answers (<i>t</i> test, $p < .05$)	34% (8.2%)	42% (6.2%)	NA	NA
	Child initiations (<i>t</i> test, $p < .005$)				
Conti-Ramsden and Friel-Patti (1984)	Mother initiations (ANOVA, $p < .01$)	0.94 (0.07)	0.86 (0.09)	0.99	0.62 (14)
	Child initiations (ANOVA, $p < .01$)	0.49 (0.14)	0.61 (0.15)	0.83	0.53 (14)
Conti-Ramsden (1990)	Mother initiations (Wilcoxon rank sum, $p < .01$)	66%	58%	No SD given	No SD given
	Complex recasts (Wilcoxon rank sum, $p < .025$)	3.6%	7.8%		
	Cohesion illocutions (Wilcoxon rank sum, $p < .01$)	52.6%	94.3%		
Cunningham, Siegel, van der Spuy, Clark, and Bow (1985)	Child interaction (ANOVA, $p < .05$; younger group)	56.2	71.2	No SD given	No SD given
	Child initiations (after noninteraction; ANOVA, $p < .001$)	23.8	55.2		
Fey, Krulik, Loeb, and Proctor-Williams (1999)	Complex recasts (<i>t</i> test, <i>ns</i>)	0.75 (0.36)	0.66 (0.35)	0.25	0.08 (10)
Proctor-Williams, Fey, and Loeb (2001)	Copula recasts at Time 1 and 3 (MANOVA, <i>ns</i>)	0.15 (0.12)	0.13 (0.13)	0.16	0.06 (10)
		0.16 (0.12)	0.12 (0.11)	0.35	0.09 (10)
Paul and Elwood (1991)	Mother expansions (a)	1.1 (2.3)	4.2 (3.4)	1.07	0.93 (28)
	percentage (<i>t</i> test, $p < .05$)	6.2 (11.4)	8.9 (6.7)	0.24	0.14 (28)
	(b) in proportion to child utterances (<i>t</i> test, <i>ns</i>)				
Rescorla and Fechnay (1996)	Mother total synchrony (<i>t</i> test, <i>ns</i>)	0.79 (0.10)	0.84 (0.13)	0.43	0.20 (18)
	Child clear verbal cues (<i>t</i> test, $p < .001$)	0.13 (0.13)	0.50 (0.19)	2.27	0.999897 (18)
Rescorla, Bascome, Lampard, and Feeny (2001)	Mother total utterances (<i>t</i> test, $p < .01$)	166.91 (53.10)	126.86 (34.45)	0.89	0.84 (32) 0.66 (21)
	Child percentage asynchronous (<i>t</i> test, $p < .05$)	22.11 (9.8)	27.48 (9.9)	0.55	0.57 (32) 0.40 (21)

Note. PLI = primary language impairment.

Effect size and power. No studies mentioned power; therefore, retrospective calculations were performed using Minitab® Version 16 (Minitab, 2013). Effect sizes were calculated using Cohen's *d*. As shown in Table 3, some studies demonstrate large effect sizes, above 0.8, supporting the existence of group differences. However, they often had low power, below the 0.8 standard, which means that caution should be taken applying these findings to a wider clinical population. Cohen's *d* will be greater among studies with smaller sample sizes, whereas studies using larger samples will be more likely to converge around smaller effect sizes.

Conclusion

The current review found issues across studies with the criteria used to define PLI, discrepancies in the severity of delay, presence of receptive delay, the level of study detail.

Methodological considerations were highlighted regarding the use of matched groups and case-control designs. Caution needs to be taken when considering the implications of results. They come from only a small number of studies, with a small cumulative number of participants, representing predominantly middle-class families in English-speaking countries, and no included studies were reported after 2001. Although some children had been referred to SLT services, they ranged in delay severity and often demonstrated expressive-only delay. These children may represent, in part, some clinical caseloads. However, children with receptive delays, or those from lower SES backgrounds, may be at greater risk of language difficulties. There is a dearth of literature with these particular subgroups, which require special attention in future research.

The review findings should be considered only as preliminary descriptive accounts. However, the review

suggests that differences in the characteristics of PCI with children with PLI compared with TD peers are limited, which challenges the idea that these two groups of children experience different communicative environments. Furthermore, differences found were generally attributed to language differences in the children, and those with PLI may learn less effectively from their environments. Examining the relationship between parent and child language behavior over time could permit analysis of factors that influence children's developmental trajectories (Tager-Flusberg, 2005), which suggests that longitudinal studies would develop understanding of the relationship between PCI and child language development. Although two studies

in the review used longitudinal designs, they did not consider how parents' language changed in relation to children's developing language skills. The influence of certain interactional characteristics may be specific to particular language or cognitive levels, which change over time (Nelson, Denninger, Bonvillian, Kaplan, & Baker, 1984; Rowe, 2012). Huttenlocher, Waterfall, Vasilyeva, Voveca, and Hedges (2010) measured parent and child language at multiple time points with TD preschoolers. More longitudinal research with children with PLI is recommended for the future to determine predictive relationships and the direction of influence between parent and child in this clinical population.

Appendix

MEDLINE Search Strategy

1. Child, Preschool/	29. vocabulary/
2. exp Infant/	30. exp Language Development/
3. (child* or infant* or toddler* or boy* or girl* or preschool age or preschool age or infancy).ti,ab.	31. ((vocab* or language or lexic* or linguist* or verbal) adj (grow* or develop* or chang* or acquisition or size or spurt or explor* or abilit*)).ti,ab.
4. Language Development Disorders/	32. ((word* adj learn*) or (early adj language)).ti,ab.
5. Language Disorders/	33. ((speech or language or vocab*) adj (express* or receptive or product* or comprehen*)).ti,ab.
6. Language Therapy/	34. #29 or #30 or #31 or #32 or #33
7. ((speech or language or communicat*) adj (delay* or disorder* or patholog* or impair*)).tw.	35. #9 and #28 and #34
8. (language develop* disorder* or late talk* or specific language impair* or SLI).tw.	36. exp Hearing Disorders/
9. #1 or #2 or #3 or #4 or #5 or #6 or #7 or #8	37. ((loss or impair) adj hear*).tw.
10. exp Parent-Child Relations/	38. (sign* language or deaf*).tw.
11. ((maternal or parent*) adj respons*) or responsiveness).ti,ab.	39. exp Intellectual Disability/
12. interact* style*.ti,ab.	40. mental retard*.tw.
13. ((maternal or parent* or mother or father) adj speech).ti,ab.	41. alternative augmentative communicat*.tw.
14. exposure.ti,ab.	42. Autistic Disorder/ or Asperger Syndrome/
15. language adj input.ti,ab.	43. Cleft Palate/
16. scaffold*.ti,ab.	44. Otitis Media/ or exp Otitis/ or Otitis Externa/ or Otitis Media with Effusion/ or Otitis Media, Suppurative/
17. ((child-direct* or child* direct* or infant-direct* or infant* direct*) adj speech).ti,ab.	45. Exp Blindness/
18. motherese.ti,ab.	46. speech disorders/ or aphasia/ or articulation disorders/ or echolalia/ or mutism/ or stuttering/
19. (parent* adj (attitude* or character* or invest* or involve* or skill* or style* or behavior* or personal*)).ti,ab.	47. exp Dyslexia/
20. Language/	48. exp Brain Damage, Chronic/
21. exp Nonverbal Communication/	49. multilingualism/
22. exp Verbal behaviour/	50. (bilingual* or second language).tw.
23. non/verbal adj communicat*.ti,ab.	51. #36 or #37 or #38 or #39 or #40 or #41 or #42 or #43 or #44 or #45 or #46 or #47 or #48 or #49 or #50
24. joint attention.ti,ab.	52. #35 and #51
25. (play behavior* or symbolic interact* or intention read* or intention*).ti,ab.	53. #35 not #52
26. ((theory adj mind) or social* cognit*).ti,ab.	
27. ((social* or environment*) adj (influenc* or interact* or language or context*)).ti,ab.	
28. #10 or #11 or #12 or #13 or #14 or #15 or #16 or #17 or #18 or #19 or #20 or #21 or #22 or #23 or #24 or #25 or #26 or #27	

Authors' Note

This review was carried out in partial fulfillment of M-level credit requirements for the degree of doctor of philosophy (PhD) at the University of the West of England (UWE), Bristol. The views expressed are those of the authors and not necessarily those of the NHS, the NIHR, or the Department of Health.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: This article summarizes independent research funded by the National Institute for Health Research (NIHR) under its Programme Grant for Applied Research Programme (RP-PG-0109-10073).

References

- Baxendale, J., & Hesketh, A. (2003). Comparison of the effectiveness of the Hanen parent programme and traditional clinic therapy. *International Journal of Language & Communication Disorders*, 38, 397–415. doi:10.1080/1368282031000121651
- Beitchman, J. H., Jiang, H., Koyama, E., Johnson, C. J., Escobar, M., Atkinson, L., . . . Vida, R. (2008). Models and determinants of vocabulary growth from kindergarten to adulthood. *Journal of Child Psychology and Psychiatry*, 49, 626–634. doi:10.1111/j.1469-7610.2008.01878.x
- Conti-Ramsden, G. (1990). Maternal recasts and other contingent replies to language-impaired children. *Journal of Speech and Hearing Disorders*, 55, 262–274. doi:10.1044/jshd.5502.262
- Conti-Ramsden, G., & Friel-Patti, S. (1983). Mothers' discourse adjustments to language-impaired and non-language-impaired children. *Journal of Speech and Hearing Disorders*, 48, 360–367. doi:10.1044/jshd.4804.360
- Conti-Ramsden, G., & Friel-Patti, S. (1984). Mother-child dialogues: A comparison of normal and language impaired children. *Journal of Communication Disorders*, 17, 19–35. doi:10.1016/0021-9924(84)90023-6
- Conti-Ramsden, G., Mok, P. L. II, Pickles, A., & Durkin, K. (2013). Adolescents with a history of specific language impairment (SLI): Strengths and difficulties in social, emotional and behavioral functioning. *Research in Developmental Disabilities*, 34, 4161–4169. doi:10.1016/j.ridd.2013.08.043
- Critical Appraisal Skills Programme. (2013). *Critical Appraisal Skills Programme: Making sense of evidence—Checklists*. Retrieved from <http://www.casp-uk.net/#/casp-tools-checklists/c18f8>
- Cunningham, C. E., Siegel, L. S., van der Spuy, H. I., Clark, M. L., & Bow, S. J. (1985). The behavioral and linguistic interactions of specifically language-delayed and normal boys with their mothers. *Child Development*, 56, 1389–1403.
- Dale, P. S., Price, T. S., Bishop, D. V. M., & Plomin, R. (2003). Outcomes of early language delay: I. predicting persistent and transient language difficulties at 3 and 4 years. *Journal of Speech, Language, and Hearing Research*, 46, 544–560.
- Fey, M. E., Cleave, P. L., & Long, S. H. (1997). Two models of grammar facilitation in children with language impairments: Phase 2. *Journal of Speech, Language, and Hearing Research*, 40, 5–19. doi:10.1044/jslhr.4001.05
- Fey, M. E., Krulik, T. E., Loeb, D. F., & Proctor-Williams, K. (1999). Sentence recast use by parents of children with typical language and children with specific language impairment. *American Journal of Speech-Language Pathology*, 8, 273–286. doi:10.1044/1058-0360.0803.273
- Gibbard, D. (1994). Parental-based intervention with pre-school language-delayed children. *International Journal of Language & Communication Disorders*, 29, 131–150.
- Higgins, J. P. T., & Green, S. (Eds.). (2011). *Cochrane handbook for systematic reviews of interventions* (Version 5.1.0). The Cochrane Collaboration. Retrieved from <http://www.cochrane.org/training/cochrane-handbook>
- Girolametto, L., Pearce, P. S., & Weitzman, E. (1996). Interactive focused stimulation for toddlers with expressive vocabulary delays. *Journal of Speech and Hearing Research*, 39, 1274–1283. doi:10.1044/jshr.3906.1274
- Gough, D. (2007). Weight of evidence: A framework for the appraisal of the quality and relevance of evidence. *Research Papers in Education*, 22, 213–228. doi:10.1080/02671520701296189
- Hart, B., & Risley, T. R. (1995). *Meaningful differences in the everyday experience of young American children*. Baltimore, MD: Paul H. Brookes.
- Hoff, F. (2003). The specificity of environmental influence: Socioeconomic status affects early vocabulary development via maternal speech. *Child Development*, 74, 1368–1378. doi:10.1111/1467-8624.00612
- Huttenlocher, J., Haight, W., Bryk, A., Seltzer, M., & Lyons, T. (1991). Early vocabulary growth: Relation to language input and gender. *Developmental Psychology*, 27, 236–248. doi:10.1037/0012-1649.27.2.236
- Huttenlocher, J., Waterfall, H., Vasilyeva, M., Vevea, J., & Hedges, L. V. (2010). Sources of variability in children's language growth. *Cognitive Psychology*, 61, 343–365. doi:10.1016/j.cogpsych.2010.08.002
- Law, J., Boyle, J., Harris, F., Harkness, A., & Nye, C. (2000). Prevalence and natural history of primary speech and language delay: Findings from a systematic review of the literature. *International Journal of Language & Communication Disorders*, 35, 165–188.
- Law, J., Garrett, Z., & Nye, C. (2003). Speech and language therapy interventions for children with primary speech and language delay or disorder. *Cochrane Database of Systematic Reviews*, (3), CD004110. doi:10.1002/14651858.CD004110
- Locke, A., Ginsborg, J., & Peers, I. (2002). Development and disadvantage: Implications for the early years and beyond. *International Journal of Language & Communication Disorders*, 37, 3–15. doi:10.1080/13682820110089911
- Mervis, C. B., & Klein-Tasman, B. (2004). Methodological issues in group-matching designs: A levels for control variable comparisons and measurement characteristics of control and target variables. *Journal of Autism and Developmental Disorders*, 34, 7–17. doi:10.1023/B:JADD.0000018069.69562.b8
- Minitab. (2013). Minitab Statistical Software. Minitab. Retrieved from <http://www.minitab.com/en-us/products/minitab/>

- National Institute for Clinical Excellence. (2004, updated 2005). *Guideline development methods: Information for national collaborating centres and guideline developers*. London, England: Author. Retrieved from http://www.nice.org.uk/niceMedia/pdf/GDM_Allchapters_0305.pdf
- Nelson, K. E., Denninger, M. S., Bonvillian, J. D., Kaplan, B. J., & Baker, N. D. (1984). Maternal input adjustments and non-adjustments as related to children's linguistic advances and to language acquisition theories. In A. D. Pellegrini & T. D. Yawkey (Eds.), *The development of oral and written language in social contexts* (pp. 31–56). Norwood, NJ: ABLEX.
- Paul, R., & Elwood, T. J. (1991). Maternal linguistic input to toddlers with slow expressive language development. *Journal of Speech and Hearing Research*, 34, 982–988.
- Pickstone, C., Goldhart, J., Marshall, J., Rees, A., & Roulstone, S. (2009). A systematic review of environmental interventions to improve child language outcomes for children with or at risk of primary language impairment. *Journal of Research in Special Educational Needs*, 9, 66–79. doi:10.1111/j.1471-3802.2009.01119.x
- Plante, E., & Swisher, L. (1993). Language matches: Illuminating or confounding? *Journal of Speech and Hearing Research*, 36, 772–776. doi:10.1044/jshr.3604.772
- Proctor-Williams, K., Fey, M. E., & Loeb, D. F. (2001). Parental recasts and production in copulas and articles by children with specific language impairment and typical language. *American Journal of Speech-Language Pathology*, 10, 155–168. doi:10.1044/1058-0360(2001)015
- Rescorla, L., Bascombe, A., Lampard, J., & Feeny, N. (2001). Conversational patterns in late talkers at age 3. *Applied Psycholinguistics*, 22, 235–251. doi:10.1017/S0142716401002053
- Rescorla, L., & Fechnay, T. (1996). Mother-child synchrony and communicative reciprocity in late-talking toddlers. *Journal of Speech and Hearing Research*, 39, 200–208. doi:10.1044/jshr.3901.200
- Roulstone, S., Wren, Y., Bakopoulou, I., Goodlad, S., & Lindsay, G. (2012). *Exploring interventions for children and young people with speech, language and communication needs: A study of practice* (Research Report No. DFE-RR247-BCRP13). London, England: Department for Education.
- Rowe, M. L. (2012). A longitudinal investigation of the role of quantity and quality of child-directed speech in vocabulary development. *Child Development*, 83, 1762–1774. doi:10.1111/j.1467-8624.2012.01805.x
- Snowling, M. J., Bishop, D. V. M., Stothard, S. E., Chipchase, B., & Kaplan, C. (2006). Psychosocial outcomes at 15 years of children with a preschool history of speech-language impairment. *Journal of Child Psychology and Psychiatry*, 47, 759–765. doi:10.1111/j.1469-7610.2006.01631.x
- St Clair, M. C., Pickles, A., Durkin, K., & Conti-Ramsden, G. (2011). A longitudinal study of behavioral, emotional and social difficulties in individuals with a history of specific language impairment (SLI). *Journal of Communication Disorders*, 44, 186–199. doi:10.1016/j.jcomdis.2010.09.004
- Stothard, S. E., Snowling, M. J., Bishop, D. V. M., Chipchase, B. B., & Kaplan, C. A. (1998). Language-impaired preschoolers: A follow-up into adolescence. *Journal of Speech, Language, and Hearing Research*, 41, 407–418. doi:10.1044/jshr.4102.407
- Tager-Flusberg, H. (2005). Designing studies to investigate the relationships between genes, environments, and developmental language disorders. *Applied Psycholinguistics*, 26, 29–39. doi:10.1017/S0142716405050046
- Whitehurst, G. J., Novak, G., & Zora, G. A. (1972). Delayed speech studied in the home. *Developmental Psychology*, 7, 169–177. doi:10.1037/h0033078
- Zimmerman, F. J., Gilkerson, J., Richards, J. A., Christakis, D. A., Xu, D., Gray, S., & Yapanel, U. (2009). Teaching by listening: The importance of adult-child conversations to language development. *Pediatrics*, 124, 342–349. doi:10.1542/peds.2008-2267

6.2.4 Study Limitations

An interesting factor that has become increasingly apparent during my ongoing experience with systematic reviews is that multiple retained papers may be reporting the same piece of research from a slightly different perspective. In paper 8, only five completely separate samples were actually represented in the nine papers. So, fewer than 250 children across the two groups were actually included in analysis. The pressure on academics to publish in volume may be contributing to this slice'n'dice approach to research, perhaps the topic of a future review, but it does make conducting high quality systematic reviews more challenging. It often takes a lot of effort to unpick the relationship between papers, and as they can be published a number of years apart, or as a sub-set of a larger study, and it often is unclear how they relate to each other.

A factor we (Anna and I) were concerned about at the start of our systematic reviews was the concept of Minimal Clinically Important Difference (MCID). The MCID was first defined by Jaeschke (1989) as *'the smallest difference in score in the domain of interest which patients perceive as beneficial and which would mandate, in the absence of troublesome side effects and excessive cost, a change in the patient's management'*. Our concern was understanding what a change in a measurement actually meant. At what point does a change in a measure (in either direction, positive or negative) suggest attention or action is required from the professional caring for the person/child? Although this was not discussed in our reviews (papers 7 and 8), I would like to take a few lines to consider some aspect of MCID, that I investigated through my reading during the work undertaken to underpin these papers.

MCID values are important in understanding observed changes, to individuals and groups. For the patients, a meaningful change may be one that reflects a reduction in symptoms, or improvement in physical ability or function. A meaningful change for the healthcare professional may indicate a need to change a treatment (Cook, 2008; Crosby, Kolotkin & Williams, 2003).

There is however, no one agreed way of calculating an MCID and no clear consensus exists regarding which methods are most suitable. An extensive review of available methods was published by Wells and colleagues who classified them into nine different methods (Wells, Beaton, Shea et al, 2001). A paper that may be of more

use to a healthcare professional, however, was written by Rai, Yazdany, et al. (2015), who undertook an evaluation of the different approaches for estimating minimal clinically important differences.

The outcome of Rai, Yazdany, et al. (2015) application of different methods on a single data set was to suggest that the MCID should be based on the context of each study. Therefore, to use the most appropriate method to calculate MCID, working closely with statisticians is highly recommended. But it is also necessary to select the appropriate outcome measures which, of course, needs patient and clinician input. Having a MCID related to Health Related Quality of Life (HRQoL) measures would enable researchers and systematic reviewers to draw conclusions about 'how much change' of QoL is important, rather than patterns of change.

6.2.5 Contribution to knowledge and autoethnographic issues

6.2.5.1 Contribution to knowledge

The findings of the systematic review question the therapists' pre-existing assumptions that communicative environments of children with language delay are different from those of typically developing children, although the evidence is from a small sample of children. Rather, it may be that children with language delay are less able to learn from their environment. The review highlighted the gap in understanding the relationship between parent and child language use during parent child interaction. The need for further, longitudinal research is apparent, including children ranging in type and severity of delay, across diverse socioeconomic backgrounds.

Appendix 18 has an overview of how the research undertaken in paper 8 was disseminated prior to the paper's publication. It also provides a list of papers where paper 8 has been cited, and this is followed by a quality appraisal of the paper.

6.2.5.2 Autoethnographic issues

It is hard to unpick what I learnt about the topic, as it so closely mapped the Child Talk work, on which I was the senior research associate. What does stand out for me is working with a junior researcher, very much at the start of their career. This was the first time that I felt that I was the touchstone; the person to go to ask process questions and to seek advice. Up to then, I had developed and led research, but had

either undertaken it on my own, or as part of a small team where we each had different areas of knowledge and each took the lead on those areas.

Helping Anna with her review gave us a concrete task to focus on and develop a work based relationship. As we were both undertaking a review for the first time (although I was working on paper 7 in parallel it was not yet published), we were peers in the process. So it was easy to have open discussions around confusions and frustrations of the process, and this helped in developing the team spirit we ended up with within the Child Talk team of RAs.

I must admit that I did initially find working with Anna and the three Child Talk Research Assistants a challenge, as I was used to doing the work myself. Handing over tasks and then being asked lots of questions seemed to take longer than just doing it myself. However, as time passed and they grew in confidence and I grew in experience, I was able to focus on my work and their development. I knew that in the long run we would all improve our skills if we worked on sharing our strengths and supporting each other. This was particularly important, as the senior members of the team had limited time to be directly involved in the day-to-day delivery of the work.

6.2.6 Future research questions

Over the duration of the Child Talk project I developed an interest in parental understanding of speech, language and communication needs. This was present in the final Child Talk report in the two factors we labelled 'Adult Understanding' and 'Adult-Child Interaction'. When thinking about these two factors, and the impact of the child's environment as covered in paper 8, I began to hypothesise what it was about a "standard" home environment that a "normal" child experiences that enhances communication and how that impacts a child with a communication need. This led to discussions with Professor Sue Roulstone, Chief Investigator of Child Talk and then the development of a bid to the Economic and Social Research Council. Although this bid was not successful, we have subsequently been awarded money from the Heather van der Lely foundation to fund a PhD studentship to undertake this investigation, and I will be a supervisor on the project.

Although open to change, the project has been conceptualised to improve understanding of the range and diversity in language and interaction environments that children with language impairment are exposed to in order to inform the theory

and practice of parent-child interactions that are used in interventions with children with LI.

The specific objectives will be to:

1. To determine the variation in linguistic and social practices in home environments of children with LI across diverse communities.
2. To understand how a range of parental ethno-theories (i.e., parents' explanations of their customs and beliefs relating to linguistic and social practices at home) impact upon language and interaction practices with children with LI.
3. To use the findings to generate intervention frameworks that are culturally and linguistically sensitive to the contexts of all families.
4. To generate techniques and strategies to support children's language development that families find acceptable and useful and that could subsequently be tested within intervention studies.
5. To understand how the methodological approach used in this research contributes to our understanding and study of LI and its interventions.

The systematic review presented as paper 8 identified the gap in knowledge that allowed me to develop this hypothesis that we will now explore more fully.

6.3 Paper 9 - Clinical experience leading to the research work and production of the paper

As mentioned in 6.2, I started working at the Bristol Speech and Language Therapy Research Unit in July 2011. My role was to be the senior research associate on the NIHR programme grant, Child Talk. As a programme grant, there were six streams of work to be undertaken; 1) Speech and Language Therapy practice, 2) Parental understanding and experience of speech and language therapy, 3) Under-served groups' experience of speech and language therapy, 4) Children's engagement with speech and language therapy, 5) Documentary analysis of speech and language therapy services and the service pathways, and 6) Systematic review.

I was the only full time member of the team working on this grant and as such acted as a hub for all activities. I was also responsible for managing the work of three RAs. At the start of the project, we had a team meeting and split the streams between us, so that each research assistant had ownership of one aspect, and a senior member of the team to work with. One of the three research assistants was a psychologist, and asked to lead the under-served groups, the other two research assistance were both speech and language therapists and selected the therapist practice and the parental understanding. The chief investigator took ownership of the documentary analysis as this would require liaison with service managers across the country, and we felt that this would be more fruitful if a senior person at least opened these discussions. This left the children's engagement and the systematic review, which I led on.

At the time, I had only really read systematic reviews. I therefore enrolled on the Masters level training mentioned previously in this chapter. The experience of running and being involved with papers 7 and 8 prepared me to undertake the much larger review that was included in Child Talk.

6.3.1 Research Questions

The aim of this study was to review systematically and critically appraise the strength of the evidence for interventions for SSD in preschool children, and then categorise those interventions that fulfilled the selection criteria within the model of classifications of interventions for SSD, devised by the authors.

6.3.2 Study Design

The systematic review reported in paper 9, is a sub-review of that undertaken as part of Child Talk. This larger review was registered with PROSPERO (reference number CRD42013006369), an international register of prospective systematic reviews.

As with papers 7 and 8, I used the Booth and Fry-Smith (2004) PICO model to guide the development of the search strategy. The ‘population’ of interest was defined as preschool children between the ages of 2 years and 5 years 11 months with Primary Speech and Language Impairment.

The Child Talk research team invested considerable time in defining ‘preschool’. Within clinical fields it has been shown that it is unlikely that a language disorder would be identified prior to the age of 2;0 (Rescorla and Schwartz, 1990; Law et al, 1998; Law et al, 2000; Broomfield and Dodd, 2004). Use of 2;0 as the earliest age also reflects the volume of evidence associating this age with accelerated language growth and increased complexity of sentences (Bauman-Waengler, 2000; Brown, 1973, McLeod and Bleile, 2003). The older age of 5;11 reflects the international average of beginning school at age 6 (Sharp, 2002). Within the UK this reaches to the end of the statutory definition of “Early Years” (the academic year in which the child turns five, EYFS, 2008). This period also covers the vast majority of typically developing children’s speech and language development including phonological development (Grunwell, 1987; Dodd & Gillon, 2001; Bauman-Waengler, 2000, McLeod and Biele, 2003), use of complex sentences, maintenance of conversation, an ability to express a variety of communicative intents and adapting their communication style dependent on the listener (Brown, 1973, James, 1990).

The papers had to include an empirical intervention, although we did not specify the nature of the intervention. A comparison group was not a requirement in the included papers but there had to be at least one outcome measurement of speech or language.

We followed the PRISMA guidelines in undertaking the review extraction and quality appraisal and then once we had finalised the retained list of articles we mapped them against the nine themes developed within the Child Talk framework of speech and language therapy practice. Figure 4 shows how the papers were spread across the themes.

Figure 4: Numbers of studies from the systematic review that were allocated to each theme

Theme	Speech	Comprehension	Expressive language	Self-monitoring	Generalisation	Foundation skills	Functional communication	Adult understanding	Adult-child interaction
Speech	33	0	5	1	17	0	0	1	1
Comprehension	0	6	5	0	3	0	0	0	2
Expressive language	5	5	28	1	10	4	5	1	8
Self-monitoring	1	0	1	2	2	1	1	0	0
Generalisation	17	3	10	2	26	2	2	0	0
Foundation skills	0	0	4	1	2	4	3	0	0
Functional communication	0	0	5	0	2	3	5	0	1
Adult understanding and empowerment	0	0	1	0	0	0	0	1	1
Adult-child interaction	1	2	8	0	0	0	1	1	9
Shading represents the total number of studies assigned to a given theme.									

Once we had identified the speech papers we further refined them against a model originally developed by first author Wren (2005). This process excluded papers that only had aspects phonological awareness and no other speech elements.

6.3.3 A systematic review and classification of interventions for speech-sound disorder in preschool children

Review

A systematic review and classification of interventions for speech-sound disorder in preschool children

Yvonne Wren^{†‡}, Sam Harding[†], Juliet Goldbart[§] and Sue Roulstone^{†¶}

[†]Bristol Speech and Language Therapy Research Unit, North Bristol NHS Trust, Bristol, UK

[‡]Faculty of Health Sciences, University of Bristol, UK

[§]Faculty of Health, Psychology and Social Care, Manchester Metropolitan University, UK

[¶]Faculty of Health and Applied Sciences, University of the West of England, Bristol, UK

(Received May 2017; accepted December 2017)

Abstract

Background: Multiple interventions have been developed to address speech sound disorder (SSD) in children. Many of these have been evaluated but the evidence for these has not been considered within a model which categorizes types of intervention. The opportunity to carry out a systematic review of interventions for SSD arose as part of a larger scale study of interventions for primary speech and language impairment in preschool children.

Aims: To review systematically the evidence for interventions for SSD in preschool children and to categorize them within a classification of interventions for SSD.

Methods & Procedures: Relevant search terms were used to identify intervention studies published up to 2012, with the following inclusion criteria: participants were aged between 2 years and 5 years, 11 months; they exhibited speech, language and communication needs; and a primary outcome measure of speech was used. Studies that met inclusion criteria were quality appraised using the single case experimental design (SCED) or PEDro-R, depending on their methodology. Those judged to be high quality were classified according to the primary focus of intervention.

Outcomes & Results: The final review included 26 studies. Case series was the most common research design. Categorization to the classification system for interventions showed that cognitive-linguistic and production approaches to intervention were the most frequently reported. The highest graded evidence was for three studies within the auditory-perceptual and integrated categories.

Conclusions & Implications: The evidence for intervention for preschool children with SSD is focused on seven out of 11 subcategories of interventions. Although all the studies included in the review were good quality as defined by quality appraisal checklists, they mostly represented lower-graded evidence. Higher-graded studies are needed to understand clearly the strength of evidence for different interventions.

Keywords: speech-sound disorder, systematic review, Child Talk, intervention.

What this paper adds

What is already known on the subject

A wide range of interventions are available for speech and language therapists to use when working with children with SSD. While some intervention approaches have robust evidence to support them, others do not have evidence or have more limited evidence.

What this paper adds to existing knowledge

This study systematically reviewed the evidence for those interventions that have been tested with children under 6 years of age. A model for classification of intervention studies in SSD is proposed and the evidence to support interventions within the model provided.

Address correspondence to: Yvonne Wren, Bristol Speech and Language Therapy Research Unit, Pines and Steps, Southmead Hospital, Westbury-on-Trym, Bristol BS10 5NB, UK; e-mail: Yvonne.wren@bristol.ac.uk

International Journal of Language & Communication Disorders
ISSN 1368-2822 print/ISSN 1460-6984 online © 2018 Royal College of Speech and Language Therapists
DOI: 10.1111/1460-6984.12371

What are the potential or actual clinical implications of this work?

Speech and language therapists will be able to identify at a glance which interventions that have been tested with children under age 6 have evidence to support them. Evidence is varied in strength and intervention studies using more robust research designs are needed to test fully the interventions described in the current literature.

Introduction

Speech-sound disorder (SSD) is a high-prevalence condition in preschool children (Broomfield and Dodd 2004, Eadie *et al.* 2015, McLeod and Harrison 2009, Shriberg *et al.* 1999). In response to this, a number of interventions have been developed that vary in the method used to achieve change in a child's speech (Baker and McLeod 2011).

To date, a number of systematic literature reviews have examined the effectiveness of these interventions for children with SSD across the age range. Some of the reviews were part of a larger and more comprehensive review of speech and language therapy interventions for children with speech and language delay or disorder (Law *et al.* 2003, 2012, 2015), while others have focused specifically on speech (Baker and McLeod 2011, Murray *et al.* 2014) or on a specific type of intervention (Lee *et al.* 2009, Lee and Gibbon 2015, McCauley *et al.* 2009, Morgan and Vogel 2008). While those focusing on specific interventions revealed a paucity of studies with sufficient strength to provide categorical support for the approaches (specifically, electropalatography, non-speech oral motor exercises and interventions for childhood apraxia of speech), the results of the more extensive reviews were encouraging. Law *et al.* (2003) included only randomized controlled trials in their review and found convincing support for interventions where the outcome was the child's 'expressive phonology'. Similarly, the review by Law *et al.* (2012) found that of 57 interventions included in the review, approximately one-third (38%) targeted speech. Evidence for most of these interventions was at a moderate level (68%), i.e., tested in either a randomized controlled trial or several quasi-experimental studies, whilst for others the evidence was at an indicative level, i.e., they have good face validity and are widely used by clinicians, but have limited research evidence that can be generalized to the population concerned.

Baker and McLeod (2011) included a wider range of study designs in their narrative review of evidence-based practice for children with SSD. Samples in these studies included participants with concomitant difficulties such as hearing loss, cleft lip and/or palate, or stuttering and spanned an age range of 1;11–10;5. They identified a total of 154 studies which described seven different methods for target selection and 46 different approaches to intervention. While a small number of

these interventions had been subject to meta-analysis or included in a randomized controlled trial, the majority had been subject to less rigorous investigations such as quasi-experimental or non-experimental case studies. Baker and McLeod concluded that more rigorous experimental design is required to enable the relative benefits of any intervention or approach to be determined.

The interpretation of Baker and McLeod's review in a clinical context is challenging. Authors of differing theories and approaches often provide clear guidance regarding the most appropriate intervention to use with children with differing presentations (e.g., Dodd and Bradford 2000). However, without comparisons of the efficacy or effectiveness of one approach over another for the full range of approaches available, clinicians are left without clear evidence of the best approach to use. This challenge is well illustrated in the 2006 special issue of *Advances in Speech-Language Pathology* on 'Jarrod', a 7-year-old boy with SSD (McLeod, 2006). This symposium published papers by different authors, who were invited to advocate and describe their own approach to intervention for this child. The different interventions were all well argued and justified at a theoretical level but not compared with each other, and there was no conclusion regarding which approach might be the most effective or efficient.

The recognition that different approaches to intervention may be needed for children with different presentations of SSD has led to a widespread call in the literature for more detailed assessment and analysis of SSD (McLeod and Baker 2004, Skahan *et al.* 2007, Stackhouse and Wells 1997). In the absence of this, clinicians tend to favour the use of just two or three named approaches, often combined into one eclectic package, presumably with the expectation that one of the elements within the package will target the child's specific needs (Joffe and Pring 2008, McLeod and Baker 2004, Roulstone *et al.* 2012). The approaches named by speech and language therapists as most frequently used often lack detail and are ambiguous in terms of how exactly they are delivered or interpreted. Terms such as 'auditory discrimination', 'meaningful minimal contrast', 'phonological awareness' (Joffe and Pring 2008), 'traditional articulation therapy' and 'minimal pairs' (McLeod and Baker 2004) and 'minimal pairs', 'auditory discrimination' and 'sequencing sounds' (Roulstone *et al.* 2015) are cited as commonly used interventions. Therefore,

it is not clear how far the approaches used frequently by clinicians map onto the approaches described in the intervention literature.

There is a need to appraise systematically the evidence for intervention in SSD and then map that onto the approaches described by clinicians. In this way, speech and language therapists with a busy and varied caseload would be more easily able to identify the strength of evidence for interventions that fit with the approach they determine is needed for an individual child.

A model for the classification of interventions for SSD

Existing classifications of SSD have focused on the child's aetiology (Shriberg *et al.* 2010), their surface-level speech presentation (Dodd 2005) or their speech-processing skills (Stackhouse and Wells 1997). A useful summary of these approaches is provided by Waring and Knight (2011). While the Dodd classification provides guidance regarding which interventions map onto each identified subtype, this only covers a small number of the range of interventions available, as identified by Baker and McLeod (2011). An alternative approach is to classify interventions and attempt to map this onto the kinds of difficulties that children with SSD might experience. This approach has been adopted in descriptions of intervention approaches by Bernthal *et al.* (2012), Rvachew and Brosseau-Lapré (2012) and Stackhouse and Wells (1997). Typically, interventions have been grouped as regards the level of processing they are primarily targeting: 'input', where the child is required to respond to some auditory stimuli to effect change in their speech; 'storage', where the child is asked to reflect on their stored representations of words as a means to challenge existing inaccurate representations; or 'output', which require the child to produce speech in response to imitation or some other stimuli.

An extension of this approach was expanded in work carried out by Wren (2005) and was used as the basis for the work carried out in the systematic review reported in this paper. Using a bottom-up approach from the intervention procedures available and identified as in use by clinicians (Roulstone and Wren 2001), the model is organized by the area where change is expected to occur in order to facilitate change in speech output. It is hypothetical and proposes one way of organizing types of intervention procedures and has changed since the original version described by Wren (2005). As such, it has the capacity to change further and evolve as new intervention procedures and new evidence become available. Nonetheless, it provides an initial framework that is inclusive of the diverse range of intervention procedures available to clinicians. Specific approaches are not named

in this model but the area where change is expected to occur and which indeed is being targeted in the intervention has been identified and categorized accordingly (figure 1).

The model labels five categories of intervention: environmental, auditory-perceptual, cognitive-linguistic, production and integrated. The environmental approach is distinct from the others in that it encompasses intervention approaches that make use of everyday interactions, rather than specific directed activities, to promote change in a child's speech-sound system. This would include procedures sometimes described as 'naturalistic intervention' as well as the modelling and recasting of a child's spontaneous productions (Camarata 2010). Auditory perceptual interventions target the child's perceptual skills as a means to induce change in speech output and include activities that aim to increase exposure to the sounds being targeted, as in focused auditory stimulation, and discrimination tasks designed to increase phoneme perception skills (Hodson and Paden 1991, Rvachew and Brosseau-Lapré 2010). Cognitive-linguistic interventions engage the child in higher-level processing in which the child's awareness of their speech is consciously addressed and used to promote change, through either confronting a child with their reduced set of contrasts or increasing awareness of sounds in speech generally. Interventions focusing on production aim to effect change through performance of oro-motor tasks, guidance on phonetic placement or manner, imitation and drills. Integrated interventions are simply those that combine two or more of the other four through profiling of the child's specific needs as in the psycholinguistic approach (Stackhouse and Wells 1997) or combining procedures into a programme of multiple interventions consistent with a 'Cycles' approach to intervention, for example (Hodson and Paden 1991).

The model does not reflect decisions around phoneme target selection, though undoubtedly the decisions regarding procedure and target are related for many interventions. Nor does it attempt to link to aetiology. However, the model makes explicit where change is expected to occur as a consequence of intervention. It is anticipated that this would provide a summary of the current evidence which is more easily accessible to clinicians, and therefore addresses some of the concerns raised by Lancaster *et al.* (2010) regarding the incompatibility of research and clinical work.

Aims

The aim of this study was to review systematically and critically appraise the strength of the evidence for interventions for SSD in preschool children and then categorize those interventions which fulfilled the selection

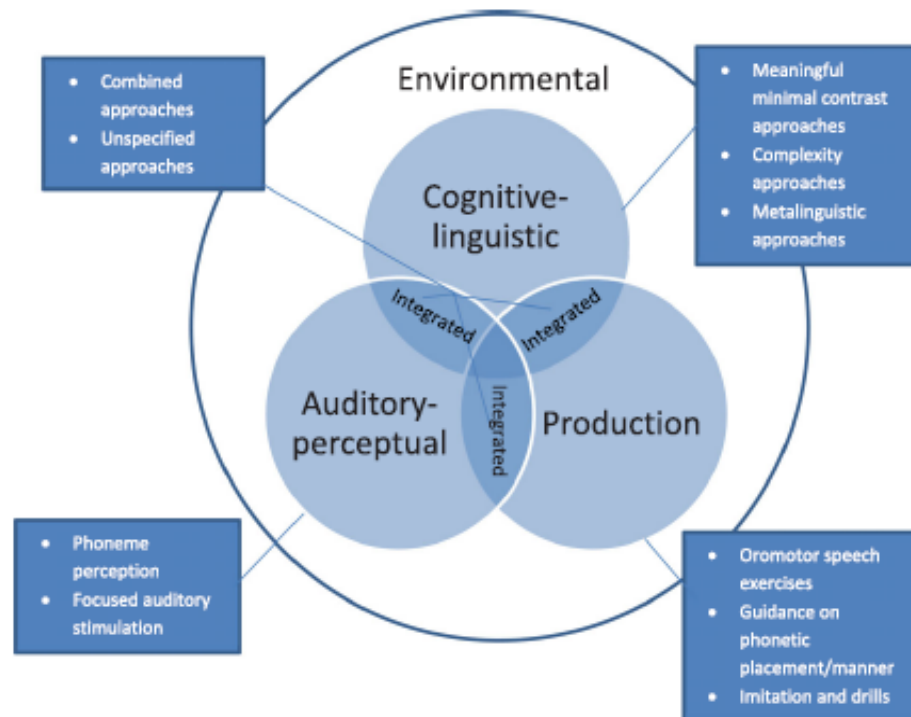


Figure 1. Model of intervention procedures for targeting speech-sound disorder (SSD). [Colour figure can be viewed at wileyonlinelibrary.com]

criteria within the model of classifications of interventions for SSD described above. Studies of interest would include children with SSD aged between 2 and 6 years; use a range of study designs; and measure outcomes in speech. The intention was that this would provide an overview of current evidence for intervention for SSD with preschool children in an easily accessible format which could be quickly be mapped onto individual children's needs.

This study was part of a larger review of interventions for children with speech and language impairment in preschool children with no concomitant difficulties (Roulstone *et al.* 2015) within the 'Child Talk' research programme, a series of research studies investigating the evidence base for speech and language therapy intervention for preschool children.

Method

The systematic review was guided by the principles outlined in the Cochrane Collaboration methodology (Higgins and Green 2011), as far as they could be applied

to the study methodologies, and built on the review undertaken by Pickstone *et al.* (2009). The search strategy described below outlines the larger review carried out for the Child Talk research programme and describes how the studies relevant to SSD were identified within this. The systematic review was registered with PROSPERO (registration number CRD42013006369), an international register of prospective systematic reviews.

Search strategy

The search strategy employed three key elements: (1) the development of a comprehensive and relevant list of search terms to ensure that all potentially valid studies in relation to interventions for speech and language impairment without concomitant difficulties were returned; (2) the exploration of a suitably broad range of databases to capture as many potentially valid studies as possible, including published, unpublished and conference proceedings; and (3) the identification of clear inclusion criteria against which to filter potentially valid studies and provide the dataset for analysis. The

authors and co-applicants of the Child Talk programme of research (Roulstone *et al.* 2015) identified a set of search terms based on their previous work in the field (Blackwell *et al.* 2014, Hambly *et al.* 2013, Marshall *et al.* 2011, Pickstone *et al.* 2009, Wren *et al.* 2013). Further potential search terms were identified from key papers. This expertise was augmented by consultation with information specialists. Through an iterative process of identification and discussion, a list of 90 search terms was determined to provide the most appropriate set to capture potentially valid studies (see appendix A). The same process was used to select appropriate databases to ensure maximum inclusion of published data, unpublished data and conference proceedings.

In line with Booth and Fry-Smith (2003), the PICO model (population, intervention, comparison, outcome) guided the development of the inclusion criteria. All research design methodologies were considered and therefore the 'comparison' element of the PICO model was not used to determine eligibility, but recorded during data extraction. For inclusion in the larger Child Talk review, studies had to meet the following requirements:

- Population: at least 80% of the sample was required to be within the age range 2;00–5;11 at the start of the intervention or at recruitment; children would be diagnosed or considered 'at risk' of speech and language impairment without concomitant difficulties.
- Intervention: an empirical evaluation of an intervention, including randomized controlled trials, experimental and quasi-experimental studies and case studies, which included multiple baseline or other systematic manipulation of the intervention.
- Outcomes: at least one of the primary outcome measures of included studies would address speech, language, communication or interaction; at a later stage, those studies that included primary outcome measures of speech were included in this topic specific review (see below).

Studies were excluded if:

- they related to children whose speech or language appeared to be developing typically with no evidence to suggest that their language was 'at risk';
- they related to children whose speech or language delays were associated with other developmental or pervasive conditions such as learning difficulties, autism, cleft palate and cerebral palsy; and/or
- the only outcomes were social or behavioural.

Search procedure

A combination of 'free text' terms with Boolean operators and truncations was used. Eighteen separate searches were conducted in electronic databases (see appendix B) to identify appropriate studies in papers published from the earliest entries of any of the databases until January 2012. Papers were initially reviewed by title and then by abstract.

Reliability

Two of the authors independently reviewed the titles of 10% of the papers identified from the initial search of the databases to screen for relevance, removing any studies that did not fit the exclusion and inclusion criteria. There was 100% consensus and the remaining 33,000 references were shared between these two authors and papers were excluded at the title level. This process led to the retention of 4574 papers. The abstract review was undertaken by four members of the research team, with two people for each manuscript (one speech and language therapist and one psychologist). Where disagreements occurred, discussion took place within the team until consensus was reached. Those papers retained at this stage were then reviewed in their entirety in light of the inclusion and exclusion criteria.

The retained papers were further reduced to those that had interventions which related to SSD. Studies were included at this stage if the intervention described in the research was consistent with the definition: 'Work that increases the accuracy of speech production or articulation, often focusing on specific sound(s).' Those studies that focused on phonological awareness skills only and did not relate to speech output were excluded. The remaining papers were then subjected to a quality appraisal.

Quality appraisal

The quality appraisal tools used in this review were selected to be relevant to the research designs used in the included studies. Two tools were used for this purpose: (1) the Physiotherapy Evidence Database quality assessment tool (PEDro-P; Perdises and Tate 2009) had a score range of 0–9 and was used to appraise the methodological quality of randomized and non-randomized controlled trials; and (2) single-case experimental design (SCED) had a score range of 0–10 and was used for single case studies (Tate *et al.* 2008). All appraisers undertook and passed training on PEDro-P and SCED.¹ Each paper was reviewed by at least two researchers, and if disagreement had occurred, it was planned to discuss and reach consensus. This process was not required as agreement on the quality assessment was 100%. For

Table 1. Process of categorization of procedures in intervention for speech-sound disorder (SSD)

	Environmental	Auditory perceptual	Cognitive linguistic	Production	Combined
Description	Procedures incorporated into everyday interactions	Procedures that target listening and perceptual skills	Procedures that require the child to reflect on their speech and/or increase their awareness of speech generally	Procedures that aim to effect change through instruction on production and production practice	Procedures that combine two or more of the other four categories into a tested intervention
Examples	Modelling, recasting	Auditory discrimination, focused auditory stimulation, phoneme perception tasks	Contrast therapy, metalinguistic tasks	Drills, guidance on phonetic placement or manner, traditional articulation	Cycles approach, psycholinguistic approach

both tools, a higher score was associated with a greater quality of the methodology applied and reported within the study. In line with previous reviews (Canarinos and Marinko 2009; Maher *et al.* 2003), a score of 6 or over was used to identify studies of acceptable quality which would be retained in the review. These studies were then mapped onto the classification of intervention procedures model described above.

Data extraction and synthesis

The process of synthesis consisted of two stages. The first stage extracted the characteristics of the studies relating to country, culture and language(s) of the researchers and participants and to study designs categorized using the National Health and Medical Research Council (NHMRC) levels of evidence guidelines (NHMRC 2007). A wide range of study designs was included in the review. This was to acknowledge that those with a lower level of evidence could be developed into trials using higher-graded designs in future.

The second stage extracted information on location and agent of intervention, assessment and outcome measures used, number of treatment sessions, and a description of the intervention provided. The description of the intervention was used to map the study onto the model of intervention procedures. Specifically, the information provided in the paper that described the procedures (as opposed to targets or the underlying theory) carried out to effect change in the child's speech sounds was considered to identify the best fit with the categories within the model described in the introduction. Where more than one type of procedure was included in the intervention protocol but only one category was under investigation, the study would be classified under the category that was the best fit for the element of the intervention being investigated. Where a combination of types of procedure had been implemented, these were noted and the study assigned to the 'integrated' category. Table 1 provides a summary of the criteria used to categorize intervention procedures described in each paper.

Subsequently, effect sizes for speech outcomes were calculated where data were available and appropriate. This was undertaken using the Campbell Collaboration effect size calculator.² Studies using a within-subject pre-post methodology providing sufficient information were assessed using a second online calculation tool³ and single-subject experimental designs were assessed using improvement rate difference (IRD; Parker *et al.* 2011).

Results

Figure 2 shows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)⁴ flowchart and summary of papers retrieved at each stage of the review. Of the 147 studies matching the inclusion criteria for the Child Talk project as a whole, 55 could be mapped onto the speech theme. Twenty-five of these papers, reporting on 26 studies, demonstrated a sufficient level of quality (i.e., obtained of 6 or more) when assessed using the PEDro-P or SCED scale. Of the 30 that did not attain a score of 6 or more on these measures, 11 were reviewed using PEDro-P and 19 with SCED. The mean average scores on these excluded studies were 4 and 3 respectively (medians of 4 and 3 respectively). The most frequent deficits in the randomized and non-randomized controlled studies were lack of concealment during group allocation and lack of blinding of the assessor who measured at least one key outcome. In the single-case experimental studies, the top three deficits in reporting were: lack of raw data being reported; assessors not being independent of treatment/intervention; and lack of replication either across subjects, therapists or setting.

Categorization of studies and reported outcomes

Of the 26 studies retained for inclusion, 18 were undertaken in the United States, four in Canada, three in Australia and one in the UK. Fifteen of the studies used a case series design and three were case studies. A further three studies used a randomized controlled trial design

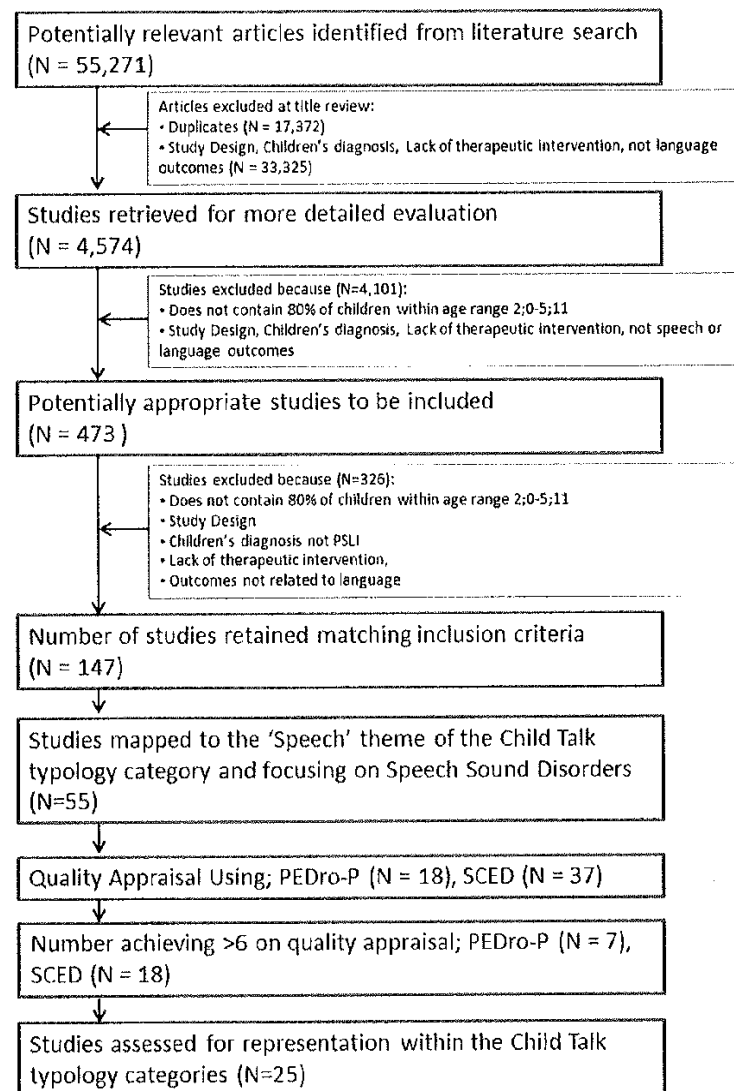


Figure 2. Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow chart. [Colour figure can be viewed at wileyonlinelibrary.com]

and a further four used a between-groups design. The 26 studies were categorized according to the procedure used in the intervention using the model in figure 3. It was possible to calculate effect sizes in 10 of the studies and to provide a range of the improvement rate difference in single cases for three more. Table 2 details

each of the studies in the review and provides summary information on each obtained from the data extraction.

Environmental approaches are represented by one study. Yoder *et al.* (2005) was categorized here due to the intervention using recasting and modelling within clinic contexts. This study found no main effect of the

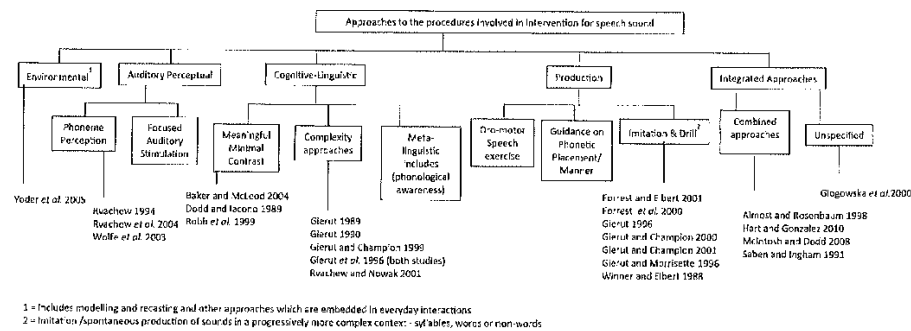


Figure 3. Evidence for intervention procedures for preschool children with speech-sound disorder (SSD). [Colour figure can be viewed at wileyonlinelibrary.com]

broad target recast intervention but did report a positive long-term impact on intelligibility for children with low pretreatment speech accuracy in comparison with standard care.

Within the category of auditory perceptual approaches, the subcategory of phoneme perception approaches was used in three studies (Rvachew 1994, Rvachew *et al.* 2004, Wolfe *et al.* 2003). The children in Rvachew (1994) were randomly allocated to three groups and these children were given listening tasks focused on treatment of misarticulated versions of target words. Rvachew *et al.* (2004) used training in phonemic perception, letter recognition, letter-sound association and onset-rime matching. Both studies found a positive effect of the intervention. In contrast, Wolfe *et al.* (2003) compared sound identification training plus production training with production-only training and found no difference between the two groups except for sounds which were poorly identified prior to intervention. None of the studies in the review was classified under the focused auditory stimulation subcategory.

Cognitive-linguistic approaches were the most commonly reported interventions within the studies in the review. These studies focused on three subcategories of intervention: 'meaningful minimal contrast' approaches, 'complexity' approaches and 'metalinguistic approaches'. Three studies focused on meaningful minimal contrast (Baker and McLeod 2004, Dodd and Iacono 1989, Robb *et al.* 1999) and a further six studies (from five papers) form the evidence base for (Gierut 1989, 1990, Gierut and Champion 1999, Gierut *et al.* 1996) and against (Rvachew and Nowak 2001) complexity approaches. These studies have small samples but suggest a positive impact of the interventions on the children, with one exception where change to the target of intervention was not observed (Gierut and Champion 1999).

No studies were included in the review under the category of metalinguistic approaches.

Studies within the review that came under the category of production were identified within the subcategories of 'oro-motor speech exercises', 'guidance on phonetic placement/manner' and 'imitations and drill'. No studies were categorized under 'oro-motor speech exercises' or 'guidance on phonetic placement/manner'. The seven studies within the 'imitations and drill' subcategory all worked on increasing the complexity of articulation in graded steps such as breaking words into constituent sounds and subsequently recombining to form the word (Forrest and Elbert 2001, Forrest *et al.* 2000, Gierut 1996, Gierut and Champion 1999, 2001, Gierut and Morrisette 1996, Winner and Elbert 1988). Five of these studies showed an improvement in the intervention group (Forrest and Elbert 2001, Forrest *et al.* 2000, Gierut and Champion 2000, 2001, Gierut and Morrisette 1996), while in two studies there was no statistical impact of the intervention on the child's speech output (Gierut 1996, Winner and Elbert 1988). It is important to note, however, that the purpose of the intervention Winner and Elbert (1988) was to investigate the impact of administering repeated probes during intervention with the intention that a desired outcome would be no change in performance on the probe measure, indicating that this approach can continue to be used in future trials of intervention for SSD.

'Integrated' approaches to intervention were represented by studies within the subcategories of 'combined' approaches and 'unspecified'. Combined approaches were adopted in four studies included in the review (Alm and Rosenbaum 1998, Hart and Gonzalez 2010, McIntosh and Dodd 2008, Saben and Ingham 1991). They used a combination of activities and strategies as interventions, described as being targeted at the individual child's needs or as routine one-to-one therapy.

Table 2. Summary of studies from the systematic review

Reference and country of origin	Number of child participants (number of children in each group, if applicable)	Age range (months)	Study design (type of evidence*)	No. of therapy sessions/agent of delivery	Length of each session (min)	Frequency of sessions	Duration of intervention	Type of speech sampled	Analysis used to measure change	PEDro-P/SCED score	Effect size Cohen <i>d</i> unless otherwise specified
<i>Environmetrics</i> Yoder <i>et al.</i> (2015), USA	52 (26, 26)	Group 1: average 44.3 Group 2: average 43.2	Randomized (type II)	Group 1: control 0; Group 2: treatment (treatment group) 77/SLP	30 min	Three times per week	6 months	Spontaneous speech	Percentage intelligible utterances PVC ^b POC ^b	PEDro-P 7	49 taken directly from the paper
<i>Acoustic perceptual phoneme perception</i> Roach (1994), Canada	27 (13, 9, 5)	Group 1: average 53.4 Group 2: average 53.6 Group 3: average 51.5	Randomized (type I)	6/SLP	45 min	Weekly	6–11 weeks	Word identification Single word naming	Percentage correct word identification Number of single word produced correctly	PEDro-P 6	0.0932
Roach <i>et al.</i> (2004), Canada	34 (17, 17)	Group 1: average 52.88 Group 2: average 50.29	Randomized (type II)	16 (in addition to their regular therapy)/SLP	15 min	Weekly	4.73 months	Conversation	POC ^b	PEDro-P 6	0.8376
Wells <i>et al.</i> (2003), USA	9 (4, 5)	Group 1: 47–55 Group 2: 41–50	Comparative studies—randomized therapy approach (type II)	Average 11/SLP	30 min	Twice weekly	One academic quarter	Probe list	Accuracy of production Sound identification	PEDro-P 5	–0.3634
<i>Cognitive-linguistic meaningful minimal contrast</i> Baker and McLeod (2004), Australia	2	Subject 1: 57 Subject 2: 52	Single-subject studies—case report, A-B, multiple baseline design (type IV)	1–12 2–52/SLP	45 min	Twice weekly	1–6 weeks 2–16 weeks	Probe conversation	Percentage correct production of trained clause	SCED 7	0.601 ^c
Dodd and Iacono (1989), Australia	7	36–57	Case series pre-post-intervention design (type IV)	3–40/SLP	n.a.	Weekly	Average 23.6 weeks	Spontaneous speech (during play)	POC ^b Phoneme inventory Process analysis SLU ^b	SCED 6	–1.362 ^c
Robb <i>et al.</i> (1999), USA	1	48	Case study single subject pre-post-intervention design (type IV)	20/SLP	45 min	Twice weekly	10 weeks	Speech sample Vowel list Probe list	Percentage accuracy Vowel inventory PVC ^b Acoustic analyses of vowels (duration, fundamental frequency)	SCED 6	Insignificant data

Continued

Table 2. Continued

Reference and country of origin	Number of child participants; number of children in each group, if applicable	Age range (months)	Study design (type of evidence)	No. of therapy sessions/agent of delivery	Length of each session (min)	Frequency of sessions	Duration of intervention	type of speech sampled	Analysis used to measure change	PEDro-P/SCED score	Effect size Cohen <i>d</i> unless otherwise specified
<i>Cognitive-linguistic complexity approaches</i>											
Gierat (1989), USA	55		Care study, pre-post-intervention design (type IV)	23/SLP	30 min	Twice weekly	1–5 weeks	Probe lists	Percentage accurate production of target phonemes	SCED 8	Insufficient data
Gierat (1990), USA	3	40–58	Alternating treatment design—multiple baseline design (type III-3)	n.a./SLP	60 min	Three times a week	n.a.	Probe list	Percentage accuracy correct on probe list	SCED 9	Figures are of insufficient resolution to extract data
Gierat and Champion (1995), USA	2	43–56	Single-subject studies—multiple baseline design (type III-3)	12/SLP	60 min	Three times per week	About 7 weeks	Probe	Percentage accuracy correct on probe list	SCED 6	Figures are of insufficient resolution to extract data
Gierat <i>et al.</i> (1996), study 1, USA	3	43–66	Single-subject studies—alternating treatment design (type III-2)	Up to 19/SLP	60 min	Three times per week	n.a.	Probe list	Percentage accuracy correct on probe list	SCED 7	Figures are of insufficient resolution to extract data
Gierat <i>et al.</i> (1996), study 2, USA	6	41–66	Single-subject studies—multiple baseline design (type III-2)	n.a./SLP	n.a.	n.a.	n.a.	Probe list	Percentage accuracy correct on probe list	SCED 7	Figures are of insufficient resolution to extract data
Rochow and Nowak (2001), Canada	48 (24, 24)	Group 1: average 51.46 Group 2: average 49.63	Randomized (type II)	12/SLP	n.a.	Weekly	12 weeks in two blocks of six	PPKTPs Conversation	PPKTPs PCC ^b	PEDro-P 6	Figures are of insufficient resolution to extract data –0.1194
<i>Production: imitation and drills</i>											
Forrest and Elbert (2001), USA	4	59–63	Single-subject studies—multiple baseline design (type III-2)	n.a./SLP	45 min	Twice weekly	n.a.	Probe list	PCC ^b for target phonemes	SCED 6	Insufficient data
Forrest <i>et al.</i> (2000), USA	10 (5, 5)	40–54	Comparative studies—therapy approach type III-3	n.a./SLP	n.a.	Fortnightly	n.a.	Probe	Percentage accuracy correct on probe	SCED 8	Insufficient data
Gierat (1995), USA	7	40–68	Single-subject studies—multiple baseline design (type III-2)	n.a./SLP	60 min	Three times per week	Average 18 weeks	Probe	Change in phonemic inventory	SCED 6	Insufficient data
Gierat and Champion (2000), USA	1	53	Single-subject studies—multiple baseline design (type III-2)	19/SLP	60 min	Three times per week	19	Probe list	Percentage accuracy correct on probe list	SCED 6	Insufficient data

Continued

Table 2. Continued

Reference and country of origin	Number of child participants (number of children in each group, if applicable)	Age range (months)	Study design (type of evidence ^a)	No. of therapy sessions/agent of delivery	Length of each session (min)	Frequency of sessions	Duration of intervention	Type of speech sampled	Analysis used to measure change	PEDro-P/SCED score	Effect size Cohen <i>d</i> unless otherwise specified
Gierut and Champion (2001), USA	8	40–75	Single-subject studies—multiple baseline design (type III-2)	n.a./SLP	60 min	Three times per week	n.a.	Probe list	Percentage accuracy correct on probe list	SCED 9	IRD ^d —between 94% and 100%
Gierut and Morrisette (1996), USA	2	4–62	Single-subject studies—multiple baseline design (type III-2)	n.a./SLP	60 min	Three times per week	Average of 16 weeks	Probes	Phoneme inventory	SCED 6	Insufficient data
Winner and Elbert (1988), USA	4	46–68	Single-subject studies—multiple baseline design (type III-2)	25/SLP	30 min	Three times per week	8 weeks	Speech sample Probe list Spontaneous speech (picture description)	Percentage correct scores of target sounds	SCED 7	IRD ^d —between 50% and 100%
<i>Integrated approaches: combined</i> Almos and Rosenbaum (1998), Canada	26 (13, 13)	33–61	Group studies—randomized (type I)	14–29/SLP	30 min	Twice weekly	7–15 weeks	GFTA ^b APP-R ^b Standardized test of single words Conversational speech	Single words Number of errors PCC ^b	PEDro-P 9	0.0064
Hart and Gonzalez (2010), USA	3	43–59	Single-subject studies—multiple baseline design (type III-2)	12/SLP	30 min	Twice a week	6 weeks	HAPP-3, 3 ^b Spontaneous speech sample	Process analysis Percentage sample correct	SCED 8	IRD ^d —between 0% and 100%
McLough and Dodd (2008), Australia	3	36–45	Single-subject pre-post-intervention design (type IV)	Between 12 and 38/SLP	30–40 min	Twice weekly	Between 6 and 19 weeks (average 12.8 weeks)	Single word naming test (DEAP) ^b phonology (subtest) Connected speech task (DEAP ^b) Repaired production of words (DEAP ^b —inconsistency subtest)	PVC ^b PCC ^b PPC ^b Percentage inconsistency	SCED 6	–42.187 ^c

Continued

Table 2. Continued

Reference and country of origin	Number of child participants (number of children in each group, if applicable)	Age range (months)	Study design (type of evidence ^a)	No. of therapy sessions/agent of delivery	Length of each session (min)	Frequency of sessions	Duration of intervention	type of speech sample	Analysis used to measure change	PEDro-PSCED score	Effect size Cohen <i>d</i> unless otherwise specified
Saben and Ingham (1991), USA	2	Subject 1: 52 Subject 2: 45	Single-subject studies—multiple baseline design (type III.2)	1–67 2–32 SLP	n.a.	n.a.	1–2 months 2–4.5 months	Probe list (spontaneous picture naming)	Percentage use of individual targeted phonemic processes	SCED 8	Insignificant data
<i>Intervent approaches: unspecified</i>											
Gregg et al. (2000), UK	159 (71, 94)	Group 1: 18–42 Group 2: 24–42	Comparative studies—randomized therapy approach (type II)	Average 6.2 h/SLP	Average of 47 min	Once a month	Average of 8.4 months	Unclear	Error rate	PEDro-P 8	0.0477

Notes: ^aNational Health and Medical Research Council (NH&MRC) (2007) defines hierarchy: designations of 'levels of evidence' according to the type of research question.
^bAPPR = Assessment of Phonological Processes; RPP = Repetition of Phonological Processes; HAPPR = Assessment of Phonological Processes; HAPPR = Assessment of Phonological Processes; HAPPR = Assessment of Phonological Processes; HAPPR = Assessment of Phonological Processes.
^cHAPP = Assessment of Phonological Processes; RPP = Repetition of Phonological Processes; HAPPR = Assessment of Phonological Processes; HAPPR = Assessment of Phonological Processes; HAPPR = Assessment of Phonological Processes.
^dPhonological knowledge profile (Gierut et al. 1987); Phonological knowledge profile (Gierut et al. 1987); Phonological knowledge profile (Gierut et al. 1987); Phonological knowledge profile (Gierut et al. 1987); Phonological knowledge profile (Gierut et al. 1987).
^eEffect size was calculated using a within-subject design and one-tailed test for significance.
^fIRD = improvement rate difference, a method of calculating the effect size for single-subject experimental designs (Dexter et al. 2011).

The studies provide mixed evidence for this approach: Almost and Rosenbaum (1998) showed a positive effect of active therapy in a group study while the remaining three studies reported case studies with varying patterns of response from individuals. Unspecified approaches were used in Glogowska *et al.* (2000) where no differences overall were found on the phonology score between control children and those receiving standard treatment. However, on a secondary outcome, a significantly greater proportion of children receiving treatment compared with the watchful waiting group improved their phonology such that they no longer satisfied the original phonology eligibility criteria for the trial.

Delivery of intervention

All studies included in the review used interventions that were delivered by speech and language therapists. Several studies did not provide information on the number and length of intervention sessions; however, where they did, the range was from three to 67 sessions lasting between 30 and 60 min.

Assessment measures used

Speech measurement in the reviewed studies was carried out for one or more of three purposes: to confirm eligibility for participation in the study; to identify targets for intervention; or to measure change in response to intervention (outcome measure). Three studies, all within the subcategory of phoneme perception approaches, also measured change in speech perception (Wolfe *et al.* 2003, Rvachew *et al.* 2004, Rvachew 1994). Speech output was collected using published assessments (Hart and Gonzalez 2010, McIntosh and Dodd 2008, Rvachew and Nowak 2001), confrontation picture-naming tasks devised for the study (Saben and Ingham 1991, Winner and Elbert 1988), and spontaneous continuous speech samples (Dodd and Iacono 1989, Hart and Gonzalez 2010, Saben and Ingham 1991, Rvachew 1994, Rvachew *et al.* 2004, Winner and Elbert 1988, Yoder *et al.* 2005). In all studies, reliability of the transcriptions was reported using point-to-point agreement for two transcribers, from between 20% and 100% of data collected. Some studies used a combination of two or three approaches to collecting speech samples. Several studies also used picture naming as part of a probe testing protocol (Baker and McLeod 2004, Forrest *et al.* 2000, Forrest and Elbert 2001, Gierut 1989, 1990, 1996, Gierut *et al.* 1999, Saben and Ingham 1991, Wolfe *et al.* 2003).

In terms of analysis of the speech samples collected, those studies that included published assessments within their assessment protocol typically used the analysis procedures which accompanied those tools. These

included process analysis (Assessment of Phonological Processes—Revised; Hodson 1986, 2004), phonemic or phonetic inventories (Productive Phonological Knowledge Profile; Gierut *et al.* 1987), percentage phonemes/consonants/vowels correct (Diagnostic Evaluation of Articulation and Phonology; Dodd *et al.* 2002, Shriberg and Kwiatkowski 1982), and accuracy of production (Goldman–Fristoe Test of Articulation; Goldman and Fristoe 2000). Where spontaneous speech samples, confrontation picture naming or probe lists were used, a number of analyses were carried out, as detailed in table 2.

Discussion

This systematic review of the literature has considered the evidence for a range of interventions for preschool children with SSD within a model in which interventions were classified based on the nature of the procedures used to effect change. In total, 55 papers were identified based on clearly defined search criteria. Following quality appraisal, 25 papers reporting 26 studies were appraised as robust enough to be included in the final review. These 26 studies were then mapped onto the model of interventions according to the description of the procedures within each paper.

Description of the review

While some previous reviews have limited their enquiry to children with phonological problems only (Baker and McLeod 2011), this review included any study that targeted increased accuracy of speech production or articulation, encompassing both phonological and speech motor interventions. This was important given the aim of synthesizing the evidence for clinicians who will be faced with a broad spectrum of children with SSD in practice (Broomfield and Dodd 2004, Shriberg *et al.* 2005).

The review included a range of research designs and did not limit itself to randomized control trials, though most were at level III of the NHMRC Evidence Hierarchy (NHMRC 2007) and, therefore, were either pseudo-randomized controlled trials or comparative studies with or without concurrent controls. Previous reviews (Law *et al.* 2003, Lee and Gibbon 2015, Morgan and Vogel 2008) have followed more restrictive criteria with regards to study design. However, in order to reflect the growing evidence base and the potential for lower-graded studies to develop into larger studies with more robust research designs, the decision was made to include studies with a lower level of evidence, as defined by the NHMRC (2007). This allowed an investigation of the current level of evidence for interventions and a clear picture regarding what is required to take the

evidence forward. As a counter to the inclusion of studies with lower-graded evidence, the quality appraisal tools were used to identify studies with the most robust operationalizations of these designs and reporting processes.

It should be noted, however, that where higher-graded study designs were used, results could shed a further light on lower-graded designs. For example, whereas the studies by Gierut (1989, 1990, 1996) showed a positive outcome for the complexity approach in single case designs, Rvachew and Nowak (2001) found that greater change was observed in children who received input following a developmental rather than a complexity approach to intervention in a higher-graded group study. Similarly, the group study carried out by Almost and Rosenbaum (1998) provides more convincing evidence for their combined approach to intervention in comparison with the case studies reported by others within this category of interventions.

The data-extraction process revealed that many studies did not report complete data regarding dosage, but where these were reported, there was a wide range in the number of sessions provided (3–67). However, there were no clear patterns to the dosage provided within the categories and subcategories of interventions. Rather, where it was reported, a wide range of number, frequency and duration of intervention sessions were offered. Lack of consistency in the provision of intervention makes it harder to compare across interventions and to determine the relative benefit of each.

With regards to measuring outcomes, a range of tools were used to assess speech output including published assessments, picture-naming tasks and spontaneous continuous speech samples. As with dosage, there were no clear patterns within the categories and subcategories with regard to outcome data collection and analysis. Thus, a narrative synthesis has been used rather than an attempt made at a meta-analysis where the measures differed widely. The exception to this was the subcategories of imitation and drill and complexity approaches which both relied heavily on probe word lists to test outcomes. However, these studies were predominantly carried out by two groups of researchers, which may explain the tendency towards the same measurement tools rather than indicating consensus across research groups in favour of any particular measure.

The model for classifications of interventions for SSD

The classification model used to classify those interventions included in the review was developed using a bottom-up approach based on interventions described by clinicians in practice (Roulstone and Wren 2001). The model proposes five main categories (environmental, auditory-perceptual, cognitive-linguistic,

production and integrated) that distinguish interventions according to where change, which will lead to improved speech output, is expected to occur. The subcategories attempt to capture more precisely what is being asked of the child in order to effect change. An exhaustive list of possibilities is not presented, however, and the model will undoubtedly evolve as new intervention procedures emerge and the evidence base grows.

Mapping the evidence to the model

Categorization of studies to the model was complex. Many of the studies included could have been categorized under the subcategory of 'combined', e.g., all three of the studies listed under auditory perceptual included production activities. However, studies were categorized according to the specific element of the intervention being investigated. Some studies added components to their interventions during the course of their study making it difficult to assess the particular contribution to outcome relative to the original aim of the study (McIntosh and Dodd 2008, Saben and Ingham 1991). Further difficulties arose concerning the amount of information regarding intervention procedures provided in the paper. With more information, it is possible that some of the studies reported would be re-categorized into a different group.

The majority of studies in the review focused on just three of the 11 subcategories of the model: imitations and drill (seven studies), meaningful minimal contrasts (three studies) and complexity (six studies). The remaining studies covered a further four categories/subcategories. Thus, no studies were identified for four of the subcategories of the model. It is possible that no evidence is available for each of these subcategories or that the evidence that is available was not robust enough to be included in the review, despite the broader inclusion criteria of this review compared with others. Rather than suggesting that those subcategories with no studies in the review are ineffective, the more accurate conclusion would be that currently there is no strong evidence to support these intervention procedures with preschool-aged children.

Some degree of supporting evidence was identified for seven of the intervention categories and subcategories in the model. These covered all the five main categories and a range of subcategories: environmental approaches; phoneme perception; guidance on phonetic placement/manner; imitations and drill; contrasts; complexity; combined and unspecified approaches. The number of quality studies varied across these subcategories, from just one each for 'environmental' and 'guidance on phonetic placement/manner' to seven for imitation and drill. Three subcategories in the model, imitations and drill, contrasts and complexity, were

supported by a number of good-quality studies, but the level of evidence represented in each of these studies is low based on the NHMRC (2007) classification of levels of evidence. Across these three subcategories of intervention procedure, the highest graded study was at level III-2: a comparative study with concurrent controls. This is comparable with a classification of indicative evidence based on the 'What Works' database of interventions (Law *et al.* 2015). The fact that there are studies with higher-grade evidence adds credence to the findings for the category or subcategory as a whole, but there is still a need for more studies using a higher level of evidence methodologies to strengthen the evidence base for these types of intervention. This fits with the findings of Baker and McLeod (2011) who commented on the need for higher levels of scientific rigour and the importance of replication research to build on the findings of lower-graded studies.

Higher-grade evidence was identified in the review for three studies: one using phoneme perception (Rvachew *et al.* 2004), one that used a combined approach (Almost and Rosenbaum 1998); and a third where the intervention procedure was unspecified (Glogowska *et al.* 2000). All three studies were randomized controlled trials with large sample sizes relative to most of the other studies (34, 26 and 26 respectively). Given that a range of interventions was used within these studies, this suggests there is agreement that a variety of approaches to intervention can be effective for children with SSD (Lancaster *et al.* 2010).

Clinical implications

The review and categorization of the studies onto the model of interventions, as illustrated in figure 3, provides an easy reference for clinicians regarding which interventions have evidence to support them. The categories of intervention can also be mapped onto the needs of individual children. For example, where assessment has shown that a child's presenting SSD is associated with problems in auditory processing, the interventions described by Wolfe *et al.* (2003) and Rvachew (1994) and Rvachew *et al.* (2004) could be useful. The descriptions in the individual papers regarding both the activities carried out and the manner of delivery, in terms of number and frequency of sessions, can assist in providing information for an evidence-based service. Similarly, if assessment reveals that a child's needs appear to be in the areas of cognitive-linguistic processing or production skill, the relevant studies in each category can be used to guide the plan for intervention. Though more comparative studies need to be completed to determine the degree to which some approaches are more effective or efficient than others within categories, the ability to identify specific approaches mapped to children

with specific needs is invaluable in the clinical context when time for considering the literature to cover a broad range of presentations for SSD is limited.

Strengths and limitations of the study

The systematic review had a specific remit to look at the evidence base related to intervention for SSD with preschool children (2;00–5;11). Studies with 20% or more of children outside the specified age range were not included. The criteria for inclusion meant that some frequently cited papers were not included in the review. The reasons for non-inclusion were most often related to the age range of the children in the sample or a low score on the quality appraisal tools used. Some studies were also excluded because the sample used in the study included children with known concomitant difficulties such as cleft palate or hearing loss or because outcomes were not reported for speech (see appendices C and D for excluded studies). Moreover, as the outcome measure needed to include speech output, the review did not include interventions that focused on prosodic skills or speech perception or other underlying speech processing skills unless these were included alongside a measurement of speech output.

Conclusions

To summarize, there is evidence to support certain types of intervention for preschool children with SSD and this is presented in a manner that has meaning and relevance to clinicians. Whilst there are more studies to support those interventions working on imitation and drill procedures or using cognitive–linguistic approaches, the stronger evidence is linked to working on phoneme perception, combined and unspecified approaches to intervention for children in the preschool age range. It is possible, of course, that evidence for interventions may vary in older children. Given the variation in findings across different study designs, it is important, nevertheless, for individual clinicians to read the papers themselves to understand how the intervention was delivered, the detailed characteristics of the children for whom the intervention was effective and what specifically was being investigated.

The work so far has been invaluable in establishing a preliminary evidence base in which different intervention types have been trialled and explored through small-scale studies. As well as providing initial evidence, these studies have enabled researchers to explore the facets of a particular approach to intervention. It has allowed for the understanding of issues relating to delivery which can inform both clinical practice and further investigations. Currently, there is a need for research activity to advance the knowledge base through the use of

higher-graded methodological studies which will provide more robust information on which approaches or combination of approaches are most suitable to use with this client group.

Acknowledgements

‘Child Talk’—What Works presents independent research commissioned by the National Institute for Health Research (NIHR) under its Programme Grants for Applied Research funding scheme (grant number RP-PG-0109-10073). The views expressed here are those of the authors and not necessarily those of the National Health Service (NHS), the NIHR or the Department of Health. Child Talk was a large programme and we are indebted to our co-applicants and members of the team for their wisdom, support and input to the programme as a whole, and to their influence on all outputs from the programme: Lydia Morgan, Naomi Parker, Rebecca Coad, Julie Marshall, Linda Lascelles, Jane Coad, Norma Daykin, Alan Emond, Jenny Moultrie, Tim Peters, Jon Pollock, Jane Powell, Ceres Fernandes and William Hollingworth. **Declaration of interest:** The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

Notes

1. See <http://speechbite.com/rating-research-quality/outline-rating-training-program/>.
2. See <https://www.campbellcollaboration.org/escalc/html/EffectSizeCalculator-SMD-main.php/>.
3. See <http://www.cognitivellexibility.org/effectsize/>.
4. See <http://www.prisma-statement.org> (accessed on 3 March 2016).

References

- ALMOST, D. and ROSENBAUM, P., 1998, Effectiveness of speech intervention for phonological disorders: a randomized controlled trial. *Developmental Medicine and Child Neurology*, **40**, 319–325.
- BAKER, E. and MCLEOD, S., 2004, Evidence-based management of phonological impairment in children. *Child Language Teaching and Therapy*, **20**, 261–285.
- BAKER, E. and MCLEOD, S. M., 2011, Evidence-based practice for children with speech sound disorder: part 1 narrative review. *Language, Speech and Hearing Services in Schools*, **42**, 102–139.
- BERNTHAL, J. E., BANKSON, N. W. and FLIPSEN, P., 2012, *Articulation and Phonological Disorders: Speech Sound Disorders in Children*, 6th edn (New York, NY: Pearson).
- BLACKWELL, A. K. M., HARDING, S., BABAYIGIT, S. and ROULSTONE, S., 2014, Characteristics of parent–child interactions: a systematic review of studies comparing children with primary language impairment and their typically developing peers. *Communication Disorders Quarterly*, **36**, 67–78.
- BOOTH, A. and FRY-SMITH, A., 2003, Developing the research question. In L. Topfer and I. Austin (eds), *Text on Health Technology Assessment (HTA) Information Resources* (National Information Center on Health Services Research and Health Care Technology) (available at: <https://www.nlm.nih.gov/archive/20060905/nichsr/hta/chapter2.html>) (accessed on 24 March 2017).
- BROOMFIELD, J. and DODD, B., 2004, Children with speech and language disability: caseload characteristics. *International Journal of Language and Communication Disorders*, **39**, 303–324.

- CAMARATA, S., 2010, Naturalistic intervention for speech intelligibility and speech accuracy. In A. L. Williams, S. McLeod and R. J. McCauley (eds), *Interventions for Speech Sound Disorders in Children* (Baltimore, MD: Paul H. Brookes), pp. 381–406.
- CAMARINOS, J. and MARINKO, L., 2009, Effectiveness of manual physical therapy for painful shoulder conditions: a systematic review. *Journal of Manual and Manipulative Therapy*, 17, 206–215.
- DODD, B., 2005, *Differential Diagnosis and Treatment of Children with Speech Disorder*, 2nd edn (Chichester: Wiley).
- DODD, B. and BRADFORD, A., 2000, A comparison of three therapy methods for children with different types of developmental phonological disorder. *International Journal of Language and Communication Disorders*, 35, 189–209.
- DODD, B. and IACONO, T., 1989, Phonological disorders in children: changes in phonological process use during treatment. *British Journal of Disorders of Communication*, 24, 333–352.
- DODD, B., ZHU, H., CROSBIE, S., HOLM, A. and OZANNE, A., 2002, *Diagnostic Evaluation of Articulation and Phonology (DEAP)* (London: Psychology Corporation).
- EADIE, P., MORGAN, A., UKOUMUNNE, O. C., TIOFARI EECEN, K., WAKE, M. and REILLY, S., 2015, Speech sound disorder at 4 years: prevalence, comorbidities, and predictors in a community cohort of children. *Developmental Medicine and Child Neurology*, 57, 578–584.
- FORREST, K. and ELBERT, M., 2001, Treatment for phonologically disordered children with variable substitution patterns. *Clinical Linguistics and Phonetics*, 15, 41–45.
- FORREST, K., ELBERT, M. and DINNSEN, D., 2000, The effect of substitution patterns on phonological treatment outcomes. *Clinical Linguistics and Phonetics*, 14, 519–531.
- GIERUT, J. A., 1989, Maximal opposition approach to phonological treatment. *Journal of Speech and Hearing Disorders*, 54, 9–19.
- GIERUT, J. A., 1990, Differential learning of phonological oppositions. *Journal of Speech and Hearing Research*, 33, 540–549.
- GIERUT, J. A., 1996, An experimental test of phonemic cyclicity. *Journal of Child Language*, 23, 81–102.
- GIERUT, J. A. and CHAMPION, A. H., 1999, Interacting error patterns and their resistance to treatment. *Clinical Linguistics and Phonetics*, 13, 421–431.
- GIERUT, J. A. and CHAMPION, A. H., 2000, Syllable onsets II: three-element clusters in phonological treatment. *Journal of Speech, Language, and Hearing Research*, 44, 886–904.
- GIERUT, J. A. and CHAMPION, A. H., 2001, Ingressive substitutions: typical or atypical phonological pattern? *Clinical Linguistics and Phonetics*, 14, 603–617.
- GIERUT, J. A., ELBERT, M. and DINNSEN, D. A., 1987, A functional analysis of phonological knowledge and generalization learning in misarticulating children. *Journal of Speech and Hearing Research*, 30, 462–479.
- GIERUT, J. A. and MORRISSETTE, M. L., 1996, Triggering a principle of phonemic acquisition. *Clinical Linguistics and Phonetics*, 10, 15–30.
- GIERUT, J. A., MORRISSETTE, M. L., HUGHES, M. T. and ROWLAND, S., 1996, Phonological treatment efficacy and developmental norms. *Language, Speech, and Hearing Services in Schools*, 27, 215–230.
- GLOGOWSKA, M., ROULSTONE, S., FENDERBY, P. and PETERS, T. J., 2000, Randomised controlled trial of community based speech and language therapy in preschool children. *British Medical Journal*, 321, 923.
- GOLDMAN, R. and FRISTOE, M., 1969, *Goldman-Fristoe Test of Articulation* (Circle Pines, Minnesota: American Guidance Service).
- GOLDMAN, R. and FRISTOE, M., 2000, *Goldman-Fristoe Test of Articulation—Second Edition (G-FTA-2)* (New York, NY: Pearson).
- HAMBLY, H., WREN, Y., McLEOD, S. and ROULSTONE, S., 2013, The influence of bilingualism on speech production: a systematic review. *International Journal of Language and Communication Disorders*, 48, 1–24.
- HART, S. and GONZALEZ, L., 2010, The effectiveness of using communication-centered intervention to facilitate phonological learning in young children. *Communication Disorders Quarterly*, 32, 13–25.
- HIGGINS, J. and GREEN, S., 2011, *Cochrane Handbook for Systematic Reviews of Interventions*, 5.1.0 edn. *Cochrane Collaboration* (available at: <http://www.cochrane.org/training/cochrane-handbook>) (accessed on 28 January 2016).
- HODSON, B. W., 1986, *The Assessment of Phonological Processes—Revised* (Austin, TX: PRO-ED).
- HODSON, B. W., 2004, *The Hodson Assessment of Phonological Processes—Revised* (Austin, TX: PRO-ED).
- HODSON, B. and PADEN, E., 1991, *Targeting Intelligible Speech: A Phonological Approach to Remediation*, 2nd edn (Austin, TX: PRO-ED).
- JOFFE, V. and PRING, T., 2008, Children with phonological problems: a survey of clinical practice. *International Journal of Language and Communication Disorders*, 43, 154–164.
- LANCASTER, G., KEUSCH, S., LEVIN, A., PRING, T. and MARTIN, S., 2010, Treating children with phonological problems: does an eclectic approach to therapy work? *International Journal of Language and Communication Disorders*, 45, 174–181.
- LAW, J., GARRETT, Z. and NYE, C., 2003, Speech and language therapy interventions for children with primary speech and language delay or disorder. *Cochrane Database Systematic Review*, Article No. CD004110 (available at: http://www.cochrane.org/CD004110/BEHAV_speech-and-language-therapy-interventions-for-children-with-primary-speech-and-language-delay-or-disorder) (accessed on 24 March 2017).
- LAW, J., LEE, W., ROULSTONE, S., WREN, Y., ZENG, B. and LINDSAY, G., 2012, *What Works: Interventions for Children and Young People with Speech, Language and Communication Needs*. No. DFE-RR24/BCRP10, Better Communication Research Programme (London: Department for Education).
- LAW, J., ROULSTONE, S. and LINDSAY, G., 2015, Integrating external evidence of intervention effectiveness with both practice and the parent perspective: development of 'What Works' for speech, language, and communication needs. *Developmental Medicine and Child Neurology*, 57, 223–228.
- LEE, A. S. Y. and GIBBON, F. E., 2015, Non-speech oral motor treatment for children with developmental speech sound disorders. *Cochrane Database of Systematic Reviews Issue 3*, Article No. CD009383 (available at: <http://onlinelibrary.wiley.com/doi/10.1002/14651858.CD009383.pub2/pdf>) (accessed on 24 March 2017).
- LEE, A. S. Y., LAW, J. and GIBBON, F. E., 2009, Electropalatography for articulation disorders associated with cleft palate. *Cochrane Database Systematic Reviews Issue 3*, Article No. CD006854 (available at: <http://onlinelibrary.wiley.com/doi/10.1002/14651858.CD006854.pub2/pdf>) (accessed on 24 March 2017).
- MAHER, C. G., SHERRINGTON, C., HERBERT, R. D., MOSELEY, A. M. and ELKINS, M., 2003, Reliability of the PEDro scale for rating quality of randomized controlled trials. *Physical Therapy*, 83, 713–721.
- MARSHALL, J., GOLDBART, J., PICKSTONE, C. and ROULSTONE, S., 2011, Application of systematic reviews in speech-and-language therapy. *International Journal of Language and Communication Disorders*, 46, 261–272.

- MCCAULEY, R. J., STRAND, E., LOF, G. L., SCHOOLING, T. and FRYMARK, T., 2009, Evidence-based systematic review: effects of nonspeech oral motor exercises on speech. *American Journal of Speech and Language Pathology*, **18**, 343–360.
- MCINTOSH, B. and DODD, B., 2008, Evaluation of core vocabulary intervention for treatment of inconsistent phonological disorder: three treatment case studies. *Child Language Teaching and Therapy*, **24**, 307–327.
- MCLEOD, S., 2006, Perspectives on a child with unintelligible speech. *Advances in Speech-Language Pathology*, **8**, 153–155.
- MCLEOD, S. and BAKER, E., 2004, Current clinical practice for children with speech impairment. In B. E. Murdoch, J. Gouzer, B.-M. Whelan and K. Docking (eds), *26th World Congress of the International Association of Logopedics and Phoniatrics* (Brisbane, QLD: University of Queensland).
- MCLEOD, S. and HARRISON, L., 2009, Epidemiology of speech and language impairment in a nationally representative sample of 4- to 5-year-old children. *Journal of Speech, Language and Hearing Research*, **52**, 1213–1229.
- MORGAN, A. T. and VOGEL, A. P., 2008, Intervention for childhood apraxia of speech. *Cochrane Database of Systematic Reviews*, **3**, Article No. CD006278 (available at: <http://onlinelibrary.wiley.com/doi/10.1002/14651858.CD006278.pub2/pdf>) (accessed on 24 March 2017).
- MURRAY, E., MCCABE, P. and BALLARD, K., 2014, A systematic review of treatment outcomes for children with childhood apraxia of speech. *American Journal of Speech-Language Pathology*, **23**, 486–504.
- NATIONAL HEALTH AND MEDICAL RESEARCH COUNCIL (NHMRC), 2007, *NHMRC Standards and Procedures for Externally Developed Guidelines* (Canberra, ACT: NHMRC) (available at: <http://www.nhmrc.gov.au/guidelines-publications/nh56>) (accessed on 24 March 2017).
- PARKER, R. I., VANNEST, K. J. and DAVIS, J. L., 2011, Effect size in single-case research: a review of nine nonoverlap techniques. *Behaviour Modification*, **35**, 303–322.
- PERDICES, M. and TATE, R. L., 2009, Single-subject designs as a tool for evidence-based clinical practice: are they unrecognised and undervalued? *Neuropsychological Rehabilitation*, **19**, 904–927.
- PICKSTONE, C., GOLDBART, J., MARSHALL, M., REES, A. and ROULSTONE, S., 2009, A systematic review of environmental interventions to improve child language outcomes for children with or at risk of primary language impairment. *Journal of Research in Special Educational Needs*, **9**, 66–79.
- ROBB, M. P., BLEILE, K. M. and YEE, S. S. L., 1999, A phonetic analysis of vowel errors during the course of treatment. *Clinical Linguistics and Phonetics*, **13**, 309–321.
- ROULSTONE, S. E., MARSHALL, J. E., POWELL, G. G., GOLDBART, J., WREN, Y. E., COAD, J., DAYKIN, N., POWELL, J., JASCELLES, L., HOLLINGWORTH, W., EMOND, A., PETERS, T. J., POLLOCK, J., FERNANDES, C., MOULTRIE, J., HARDING, S., MORGAN, L., HAMBLY, L., PARKER, N. and COAD, R. A., 2015, Evidence-based intervention for preschool children with primary speech and language impairments: Child Talk—an exploratory mixed-methods study. *Programme Grants for Applied Research*, **3**(5) (available at: <https://www.ncbi.nlm.nih.gov/pubmed/26312364>) (accessed on 24 March 2017).
- ROULSTONE, S. E. and WREN, Y. E., 2001, Investigation of theoretical models and therapy activities: phonological difficulties. *International Journal of Language and Communication Disorders*, **36**(Suppl.), 441–446.
- ROULSTONE, S., WREN, Y., BAKOPOULOU, I., GOODLAD, S. and LINDSAY, G., 2012, *Exploring Interventions for Children and Young People with Speech, Language and Communication Needs: A Study of Practice*. No. DFE-RR247-BCRP13 (London: Department for Education) (available at: https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/219627/DFE-RR247-BCRP13.pdf) (accessed on 24 March 2017).
- RVACHEW, S., 1994, Speech perception training can facilitate sound production learning. *Journal of Speech and Hearing Research*, **37**, 347–357.
- RVACHEW, S. and BROUSSEAU-LAPRÉ, E., 2010, Speech perception intervention. In A. I. Williams, S. McLeod and R. J. McCauley (eds), *Interventions for Speech Sound Disorders in Children* (Baltimore, MD: Paul H. Brookes), pp. 295–314.
- RVACHEW, S. and BROUSSEAU-LAPRÉ, E., 2012, *Developmental Phonological Disorders Foundation of Clinical Practice* (San Diego, CA: Plural).
- RVACHEW, S. and NOWAK, M., 2001, The effect of target-selection strategy on phonological learning. *Journal of Speech, Language, and Hearing Research*, **44**, 610–623.
- RVACHEW, S., NOWAK, M. and CLOUTIER, G., 2004, Effect of phonemic perception training on the speech production and phonological awareness skills of children with expressive phonological delay. *American Journal of Speech-Language Pathology*, **13**, 250–263.
- SABEN, C. B. and INGHAM, J. C., 1991, The effects of minimal pairs treatment on the speech-sound production of two children with phonologic disorders. *Journal of Speech and Hearing Research*, **34**, 1023–1040.
- SHRIBERG, L. D., FOURAKIS, M., HALL, S. D., KARLSSON, H. B., LORAEIER, H. L., MCSWEENEY, J. L., POTTER, N., SCHEER-COHEN, A., STRAND, E., TELKENS, C. and WISON, D., 2010, Extensions to the Speech Disorders Classification System (SDCS). *Clinical Linguistics and Phonetics*, **24**, 795–824.
- SHRIBERG, L. D. and KWIAKOWSKI, J., 1982, Phonological Disorders III: a procedure for assessing severity of Involvement. *Journal of Speech and Hearing Research*, **47**, 256–270.
- SHRIBERG, L. D., LEWIS, B. L., TOMBLIN, J. B., MCSWEENEY, J. L., KARLSSON, H. B. and SCHEER, A. R., 2005, Toward diagnostic and phenotype markers for genetically transmitted speech delay. *Journal of Speech, Language and Hearing Research*, **48**, 834–852.
- SHRIBERG, L. D., TOMBLIN, J. B. and MCSWEENEY, J. L., 1999, Prevalence of speech delay in 6-year-old children and comorbidity with language impairment. *Journal of Speech, Language and Hearing Research*, **42**, 1461–1481.
- SKAHAN, S., WATSON, M. and LOF, G., 2007, Speech-language pathologists' assessment practices for children with suspected speech sound disorder: results of a national survey. *American Journal of Speech-Language Pathology*, **16**, 246–259.
- STACKHOUSE, J. and WELLS, B., 1997, *Children's Speech and Literacy Difficulties: A Psycholinguistic Framework* (London: Whurr).
- TATE, R. L., McDONALD, S., PERDICES, M., TUGHER, L., SCHULTZ, R. and SAVAGE, S., 2008, Rating the methodological quality of single-subject designs and *n*-of-1 trials: introducing the single-case experimental design (SCED) scale. *Neuropsychological Rehabilitation*, **18**, 385–401.
- WARING, R. and KNIGHT, R., 2011, How should children with speech sound disorders be classified? A review and critical evaluation of current classification systems. *International Journal of Language and Communication Disorders*, **48**, 25–40.
- WINNER, M. and ELBERT, M., 1988, Evaluating the treatment effect of repeated probes. *Journal of Speech and Hearing Disorders*, **53**, 211–218.
- WOLFE, V., PRESLEY, C. and MESAKIS, J., 2003, The importance of sound identification training in phonological intervention. *American Journal of Speech-Language Pathology*, **12**, 282–288.

- WREN, Y., 2005, An evaluation of the use of computers in phonology therapy. Unpublished PhD thesis.
- WREN, Y., HAMBLIN, H. and ROULSTONE, S., 2013, A review of the impact of bilingualism on the development of phonemic awareness skills in children with typical speech development. *Child Language Teaching and Therapy*, 29, 11–25.
- YODER, P., CAMARATA, S. and GARDNER, E., 2005, Treatment effects on speech intelligibility and length of utterance in children with specific language and intelligibility impairments. *Journal of Early Intervention*, 28, 34–4.

Appendix A: Search terms used in a systematic review of interventions for speech-sound disorder (SSD) in preschool children

1. exp Pediatrics/
2. exp CHILD/
3. exp INFANT/
4. child\$.mp. [mp = title, original title, abstract, name of substance word, subject heading word]
5. infant\$.mp. [mp = title, original title, abstract, name of substance word, subject heading word]
6. (paediatric\$ or pediatric\$).mp. [mp = title, original title, abstract, name of substance word, subject heading word]
7. toddler\$.mp. [mp = title, original title, abstract, name of substance word, subject heading word]
8. boy\$.ti,ab.
9. girl\$.ti,ab.
10. (school child\$ or schoolchildren\$).ti,ab.
11. (pre school\$ or preschool\$).ti,ab.
12. or/1-11
13. speech disorder\$.ti,ab.
14. speech intelligibility\$.ti,ab.
15. speech therap\$.ti,ab.
16. language therap\$.ti,ab.
17. speech development.ti,ab.
18. speech delay.ti,ab.
19. language disorder\$.ti,ab.
20. language development disorder\$.ti,ab.
21. sign language\$.ti,ab.
22. child\$ language.ti,ab.
23. language therap\$.ti,ab.
24. language development.ti,ab.
25. language delay.ti,ab.
26. nonverbal communication.ti,ab.
27. non verbal communication.ti,ab.
28. communication development.ti,ab.
29. exp Speech Disorders/
30. speech Intelligibility/
31. 'rehabilitation of speech and language disorders'/or language therapy/or speech therapy/
32. Language Development Disorders/
33. Language Disorders/
34. Sign Language/
35. Child Language/
36. Language Development/
37. exp Nonverbal Communication/
38. Communication Disorders/
39. maternal responsiveness.tw.
40. directiveness.tw.
41. maternal interactive styles.tw.
42. compliance.tw.
43. maternal personality.tw.
44. child temperament.tw.
45. or/13-44
46. exp Mental Retardation/
47. exp child development disorders, pervasive/or asperger syndrome/
48. Cleft Palate/or Cleft Lip/
49. Otitis Media with Effusion/
50. exp Hearing Loss/
51. exp Blindness/
52. Stuttering/
53. Aphonia/
54. exp Pain/
55. Crying/
56. exp Analgesia/
57. Reading/
58. exp Dyslexia/
59. Cerebral Palsy/
60. (alternative and augmentative communication).mp. [mp = title, original title, abstract, name of substance word, subject heading word]
61. 'alternative and augmentative communication'.mp. [mp = title, original title, abstract, name of substance word, subject heading word]
62. exp aged/
63. geriatrics/
64. or/46-63
65. (12 and 45) not 64
66. randomized controlled trial.pt.
67. controlled clinical trial.pt.
68. randomized controlled trials/
69. random allocation/
70. double blind method/
71. single blind method/
72. clinical trial.pt.
73. exp clinical trials/
74. (clin\$ adj25 trial\$).tw.
75. ((singl\$ or doubl\$ or trebl\$ or tripl\$) adj25 (blind\$ or mask\$)).tw.
76. placebos/
77. placebo\$.tw.
78. random\$.tw.
79. research design/
80. 'comparative study'/
81. exp evaluation studies/
82. follow-up studies/
83. prospective studies/
84. (control\$ or prospectiv\$ or volunteer\$).tw.

85. (control\$ or prospectiv\$ or volunteer\$).tw.
 86. oc/66-85
 87. 'animal'/
88. 'human'/
 89. 87 not 88
 90. 86 not 89

Appendix B: Databases searched, number of results and search date

Database ^a	Search interface	Search results	Search date
MEDLINE	Ovid	8374	6 December 2011
EMBASE	Ovid	9663	6 December 2011
Cumulative Index to Nursing and Allied Health Literature (CINAHL)	EBSCOhost	8976	2 December 2011
PsycINFO	EBSCOhost	9107	11 January 2011
Cochrane Database of Systematic Reviews (CDSR)	The Cochrane Library	255	13 January 2012
Database of Abstracts of Reviews of Effects (DARE)	The Cochrane Library	0	13 January 2012
NHS Health Technology Assessment database	The Cochrane Library	0	13 January 2012
Cochrane Central Register of Controlled Trials (CENTRAL)	The Cochrane Library	0	13 January 2012
Science Citation Index	Web of Knowledge	5787	13 January 2012
Social Science Citation Index	Web of Knowledge	0	13 January 2012
International Bibliography for the Social Sciences	ProQuest	0	25 November 2011
Applied Social Sciences Index and Abstracts (ASSIA)	ProQuest	1799	25 November 2011
Sociological Abstracts ProQuest 3800 25 November 2011	ProQuest	3800	25 November 2011
Social Services Abstracts ProQuest 0 25 November 2011	ProQuest	0	25 November 2011
Educational Resource Information Center (ERIC)	ProQuest	4000	26 January 2012
Linguistics and Language Behavior Abstracts	ProQuest	3006	20 January 2012
British Education Index	ProQuest	464	20 January 2012
The Campbell Collaboration	www.campbellcollaboration.org/	40	13 January 2012

Note: ^aDatabases were searched from the date of inception to the search date.

Reference	Reason for exclusion
Alant, J. and Jager, G., 1983, Integrated speech and language therapy in the pre-school class in the special school setting. <i>Rehabilitation in South Africa</i> , 27(2), 49–53	Description of project. Not a research paper
Aram, D. M., Morris, R. and Hall, N. E., 1992, The validity of discrepancy criteria for identifying children with developmental language disorders. <i>Journal of Learning Disabilities</i> , 25(9), 549–554	Observational study
Baker, E., 2010, The experience of discharging children from phonological intervention. <i>International Journal of Speech–Language Pathology</i> , 12(4), 325–328	Discussion paper
Baker, E., 2006, Management of speech impairment in children: the journey so far and the road ahead. <i>Advances in Speech–Language Pathology</i> , 8(3), 156–163	Discussion paper
Baker, E. and McCabe, P., 2010, The potential contribution of communication breakdown and repair in phonological intervention. <i>Canadian Journal of Speech–Language Pathology and Audiology/Revue canadienne d'orthophonie et d'audiologie</i> , 34(3), 193–205	Review and discussion paper
Befi-Lopes, D. M. and Rondon, S., 2010, Syllable deletion in spontaneous speech of children with specific language impairment. <i>Pro-Fono: Revista de Atualização Científica</i> , 22(3), 333–338	Observational study
Bernhardt, B. H., Stemberger, J. P. and Major, E., 2006, General and nonlinear phonological intervention perspectives for a child with a resistant phonological impairment. <i>Advances in Speech–Language Pathology</i> , 8(3), 190–206	Discussion paper
Blacklin, J. and Crais, E. R., 1997, A treatment protocol for young children at risk for severe expressive output disorders. <i>Seminars in Speech and Language</i> , 18(3), 213–31.	Discussion paper
Bland, I. F. and Prelock, P. A., 1995, Effects of collaboration on language performance. <i>Communication Disorders Quarterly</i> , 17(2), 31–37	Participants too old
Bowen, C. and Cupples, L., 2004, The role of families in optimizing phonological therapy outcomes. <i>Child Language Teaching and Therapy</i> , 20(3), 245–260	Discussion paper
Boyle, J., 2011, Speech and language delays in preschool children. <i>British Medical Journal</i> , 343, d5181	Editorial
Browning, E., 1981, The health visitor and speech impaired children. <i>Health Visitor</i> , 54, 204–205	Discussion paper
Carter, P. and S. Edwards., 2004, EPG therapy for children with longstanding speech disorders: predictions and outcomes. <i>Clinical Linguistics and Phonetics</i> , 18(6), 359–372	Participants too old
Castiglia, P. T., 1987, Speech–language development. <i>Journal of Pediatric Healthcare</i> , 1(3), 165–167	Discussion paper

Continued

Reference	Reason for exclusion
Chang, J. Y., 2004, Case study on a profound speech-delayed subject: a behavioural approach and its implications. <i>Asia Pacific Journal of Speech, Language and Hearing</i> , 9(1), 48–53	Could not exclude language delay as part of other developmental or biological disorder
Cox, J. and Hill, S., 1993, Tackling language delay: a group work approach. <i>Health Visitor</i> , 66(8), 291–292	Discussion paper
Daly, D. A., Cantrell, R. P., Cantrell, M. L. and Aman, L. A., 1972, Structuring speech therapy contingencies with an oral apraxic child. <i>Journal of Speech and Hearing Disorders</i> , 37(1), 22–32	Participant too old
Danger, S. and Landreth, G., 2005, Child-centered group play therapy with children with speech difficulties. <i>International Journal of Play Therapy</i> , 14(1), 81–102	Unable to establish if participants are an appropriate age
Denne, M., Langdown, N., Pring, T. and Roy, P., 2005, Treating children with expressive phonological disorders: does phonological awareness therapy work in the clinic? <i>International Journal of Language and Communication Disorders</i> , 40(4), 493–504	More than 20% of the participants were too old
Dodd, B., Crosbie, S., McIntosh, B., Holm, A., Harvey, C., Liddy, M., Fontyne, K., Pinchin, B. and Rigby, H., 2008, The impact of selecting different contrasts in phonological therapy. <i>International Journal of Speech-Language Pathology</i> 10(5), 334–345	More than 20% of the participants were too old
Forrest, K. and Iuzzini, J., 2008, A comparison of oral motor and production training for children with speech sound disorders. <i>Seminars in Speech and Language</i> , 29(4), 304–311	More than 20% of the participants were too old
Gierut, J. A., 1998, Natural domains of cyclicity in phonological acquisition. <i>Clinical Linguistics and Phonetics</i> , 12(6), 481–499	Age unclear, more than 20% likely to be too old. Outcome measure phonology only
Gierut, J. A., Morrisette, M. L. and Ziemer, S. M., 2010, Nonwords and generalization in children with phonological disorders. <i>American Journal of Speech-Language Pathology</i> , 19(2), 167–177	More than 20% of the participants were too old
Gillon, G. T., 2000, The efficacy of phonological awareness intervention for children with spoken language impairment. <i>Language, Speech and Hearing Services in Schools</i> , 31(2), 126–141	More than 20% of the participants were too old
Gillon, G. T., 2002, Follow-up study investigating the benefits of phonological awareness intervention for children with spoken language impairment. <i>International Journal of Language and Communication Disorders</i> , 37(4), 381–400	More than 20% of the participants were too old
Keske-Soares, M., Brancalioni, A. R., Marini, C., Pagliarin, K. C. and Ceron, M. I., 2008, Therapy effectiveness for phonological disorders with different therapeutic approaches. <i>Pro-Fono</i> , 20(3), 153–158	More than 20% of the participants were too old
King, A. M., 2011, An integrated multimodal intervention approach to support speech and language development in children with severe speech impairments. Dissertation in ProQuest Information and Learning.	More than 20% of the participants were too old
Lanc, K. L., Fletcher, T., Carter, E. W., Dejud, C. and DeLorenzo, J., 2007, Paraprofessional-led phonological awareness training with youngsters at risk for reading and behavioral concerns. <i>Remedial and Special Education</i> , 28(5), 266–276	Participants were too old
Mire, S. P. and Montgomery, J. K., 2009, Early intervening for students with speech sound disorders. Lessons from a school district. <i>Communication Disorders Quarterly</i> , 30(3), 155–166	Review paper
Pascoe, M., Stackhouse, J. and Wells, B., 2005, Phonological therapy within a psycholinguistic framework: promoting change in a child with persisting speech difficulties. <i>International Journal of Language and Communication Disorders</i> , 40(2), 189–220	Case study: child was too old
Robertson, S. B. and Weismer, S. E., 1999, Effects of treatment on linguistic and social skills in toddlers with delayed language development. <i>Journal of Speech, Language and Hearing Research</i> , 42(5), 1234–1248	More than 20% of the participants were too young
Roulstone, S., Glogowska, M., Peters, T. J. and Enderby, P., 2004, Building good practice: lessons from a multimethod study of speech and language therapy . . . including commentary by A. A. Tyler. <i>International Journal of Therapy and Rehabilitation</i> , 11(5), 199–205	Discussion paper
Rousseau, I., Packman, A., Onslow, M., Harrison, E. and Jones, M., 2007, An investigation of language and phonological development and the responsiveness of preschool age children to the Lidcombe Program. <i>Journal of Communication Disorders</i> , 40(5), 382–397	Stuttering
Ruder, K. E. and Buncer, B. H., 1981, Articulation therapy using distinctive feature analysis to structure the training program: two case studies. <i>Journal of Speech and Hearing Disorders</i> , 46(1), 59–65	Review paper with a case study without relevant assessments
Ruscello, D. M., 1995, Visual feedback in treatment of residual phonological disorders. <i>Journal of Communication Disorders</i> , 28(4), 279–302.	Discussion paper
Ruscello, D. M., Yanero, D. and Ghalichebaf, M., 1995, Cooperative service delivery between a university clinic and a school system. <i>Language, Speech and Hearing Services in Schools</i> , 26(3), 273–277	Case study child too old
Segers, E., Nooijen, M. and de Moor, J., 2006, Computer vocabulary training in kindergarten children with special needs. <i>International Journal of Rehabilitation Research</i> , 29(4), 343–345	More than 20% of the participants were too old

Continued

Reference	Reason for exclusion
Segers, E. and Verhoeven, L., 2005, Effects of lengthening the speech signal on auditory word discrimination in kindergartners with SLI. <i>Journal of Communication Disorders</i> , 38(6), 499–514	More than 20% of the participants were too old
Sharp, F. M. and Hillenbrand, K., 2008, Speech and language development and disorders in children. <i>Pediatric Clinics of North America</i> , 55(5), 1159–1173.	Discussion paper
Shiller, D. M., Rvachew, S. and Brossseau-Lapr�, E., 2010, Importance of the auditory perceptual target to the achievement of speech production accuracy. <i>Canadian Journal of Speech–Language Pathology and Audiology/Revue canadienne d’orthophonie et d’audiologie</i> , 34(3), 181–192	Case study 1: child too old. Case study 2: English was a second language
Simsen, J. L., 1993, Auditory–verbal intervention: infants and toddlers. <i>Volta Review</i> , 95(3), 217–229	Discussion paper
Skau, L. and Cascella, P. W., 2006, Using assistive technology to foster speech and language skills at home and in preschool. <i>Teaching Exceptional Children</i> , 38(6), 12–16.	Discussion paper
Skelton, S. L., 2004, Concurrent task sequencing in single-phoneme phonologic treatment and generalization. <i>Journal of Communication Disorders</i> , 37(2), 131–155	Children were too old
Skelton, S. L. and Funk, T. E., 2004, Teaching speech sounds to young children using randomly ordered, variably complex task sequences. <i>Perceptual and Motor Skills</i> , 99(2), 602–604	Cannot exclude the possibility that impairment is due to other developmental disorder or pervasive condition
Stackhouse, J., Wells, B., Pascoe, M. and Rees, R., 2002, From phonological therapy to phonological awareness. <i>Seminars in Speech and Language</i> , 23(1), 27–42.	Review paper
Torgesen, J. K. and Davis, C., 1996, Individual difference variables that predict response to training in phonological awareness. <i>Journal of Experimental Child Psychology</i> , 63(1), 1–21	Children were ‘at risk’ of impairment
Tyler, A. A., 2002, Language-based intervention for phonological disorders. <i>Seminars in Speech and Language</i> , 23(1), 69–81	Review paper with a case study example
Tyler, A. A., 2008, What works: evidence-based intervention for children with speech sound disorders. <i>Seminars in Speech and Language</i> , 29(4), 320–330	Review paper
Tyler, A. A., Lewis, K. E., Haskill, A. and Tolbert, L. C., 2002, Clinical Forum. Efficacy and cross-domain effects of a morphosyntax and a phonology intervention. <i>Language, Speech and Hearing Services in Schools</i> , 33(1), 52–66	No speech measures
Vicsi, K., Kovacs-Vass, E. and Barczikay, P., 1994, A speech improvement technique based on visual feedback. <i>Acta Linguistica Hungarica</i> , 42(1–2), 93–101	Review paper
Warrick, N. and Rubin, H., 1992, Phonological awareness: normally developing and language delayed children. <i>Journal of Speech–Language Pathology and Audiology/Revue d’orthophonie et d’audiologie</i> , 16(1), 11–20	Only phonological awareness measures
Willens, S. G., Lombardino, L. L., MacDonald, J. D. and Owens, R. E., 1982, Total Communication: Clinical report on a parent-based language training program. <i>Education and Training of the Mentally Retarded</i> , 17(4), 293–298	More than 20% of the participants were too old
Williams, A. L., 2005, Assessment, target selection, and intervention: dynamic interactions within a systemic perspective. <i>Topics in Language Disorders</i> , 25(3), 231–242	More than 20% of the participants were too old
Williams, A. L., 2000, Clinical Focus. Multiple oppositions: case studies of variables in phonological intervention. <i>American Journal of Speech–Language Pathology</i> , 9(4), 289–299	More than 20% of the participants were too old
Wren, Y. and Roulstone, S., 2008, A comparison between computer and tabletop delivery of phonology therapy. <i>International Journal of Speech–Language Pathology</i> , 10(5), 346–363	More than 20% of the participants were too old
Zhang, X. and Tomblin, J. B., 2000, The association of intervention receipt with speech–language profiles and social–demographic variables. <i>American Journal of Speech–Language Pathology</i> , 9(4), 345–357	More than 20% of the participants were too old

Appendix C: Studies excluded at the quality appraisal phase

Adams, C., Nightingale, C., Hesketh, A. and Hall, R., 2000, Targeting metaphonological ability in intervention for children with developmental phonological disorders. *Child Language Teaching and Therapy*, 16, 285–299.

Baker, E. and Bernhardt, B., 2004, From hindsight to foresight: working around barriers to success in phonological intervention. *Child Language Teaching and Therapy*, 20, 287–318.

Bowen, C. and Cupples, L., 1998, A tested phonological therapy in practice. *Child Language Teaching and Therapy*, 14, 29–50.

Bryan, A. and Howard, D., 1992, Frozen phonology thawed: the analysis and remediation of a developmental disorder of real word phonology. *European Journal of Disorders of Communication*, 27, 343–365.

Dodd, B. and Barker, R., 1990, The efficacy of utilizing parents and teachers as agents of therapy for children with phonological disorders. *Australian Journal of Human Communication Disorders*, 18, 29–45.

Eiserman, W. D., Weber, C. and McCoun, M., 1995, Parent and professional roles in early intervention. A longitudinal comparison of the effects of two intervention configurations. *Journal of Special Education*, 29, 20–44.

- Fey, M. E., 1993, Two approaches to the facilitation of grammar in children with language impairment: an experimental evaluation. *Journal of Speech and Hearing Research*, **36**, 141–157.
- Fey, M. E., Cleave, P. L. and Long, S. H., 1997, Two models of grammar facilitation in children with language impairments: phase 2. *Journal of Speech, Language and Hearing Research*, **40**, 5–19.
- Fey, M. E., Cleave, P. L., Ravida, A. I., Long, S. H., Dejmaj, A. E. and Easton, D. L., 1994, Effects of grammar facilitation on the phonological performance of children with speech and language impairments. *Journal of Speech and Hearing Research*, **37**, 594–607.
- Gierut, J. A., 1998, Production, conceptualization and change in distinctive featural categories. *Journal of Child Language*, **25**, 321–341.
- Gillon, G. T., 2005, Facilitating phoneme awareness development in 3- and 4-year-old children with speech impairment. *Language, Speech, and Hearing Services in Schools*, **36**, 308–324.
- Grawburg, M. and Rvachew, S., 2007, Phonological awareness intervention for preschoolers with speech and sound disorders. *Canadian Journal of Speech–Language Pathology and Audiology/Revue canadienne d'orthophonie et d'audiologie*, **31**, 19–26.
- Hesketh, A., Adams, C., Nightingale, C. and Hall, R., 2000, Phonological awareness therapy and articulatory training approaches for children with phonological disorders: a comparative outcome study. *International Journal of Language and Communication Disorders*, **35**, 337–354.
- Hoffman, P. R., Norris, J. A. and Monjure, J., 1990, Comparison of process targeting and whole language treatments for phonologically delayed preschool children. *Language, Speech, and Hearing Services in Schools*, **21**, 102–109.
- Major, E. M. and Bernhardt, B. H., 1998, Metaphonological skills of children with phonological disorders before and after phonological and metaphonological intervention. *International Journal of Language and Communication Disorders*, **33**, 413–444.
- Montgomery, J. K. and Bonderman, I. R., 1989, Serving preschool children with severe phonological disorders. *Language, Speech, and Hearing Services in Schools*, **20**, 76–84.
- Ruscello, D. M., Cartwright, L. R., Haines, K. B. and Shuster, L. I., 1993, The use of different service delivery models for children with phonological disorders. *Journal of Communication Disorders*, **26**, 193–203.
- Rvachew, S., Rafaat, S. and Martin, M., 1999, Stimulability, speech perception skills, and the treatment of phonological disorders. *American Journal of Speech–Language Pathology*, **8**, 33–43.
- Shriberg, L. D., Kwiatkowski, J. and Snyder, T., 1989, Tablet versus microcomputer-assisted speech management: stabilization phase. *Journal of Speech and Hearing Disorders*, **54**, 233–248.
- Stoel-Gammon, C., Stone-Goldman, J. and Glaspey, A., 2002, Pattern-based approaches to phonological therapy. *Seminars in Speech and Language*, **23**, 3–13.
- Tyler, A. A., Edwards, M. L. and Saxman, J. H., 1987, Clinical application of two phonologically based treatment procedures. *Journal of Speech and Hearing Disorders*, **52**, 393–409.
- Tyler, A. A., Edwards, M. L. and Saxman, J. H., 1990, Acoustic validation of phonological knowledge and its relationship to treatment. *Journal of Speech and Hearing Disorders*, **55**, 251–261.
- Tyler, A. A., Gillon, G., Macrae, T. and Johnson, R. L., 2011, Direct and indirect effects of stimulating phoneme awareness vs. other linguistic skills in preschoolers with co-occurring speech and language impairments. *Topics in Language Disorders*, **31**, 128–144.
- Tyler, A. A. and Lewis, K. E., 2005, Relationships among consistency/variability and other phonological measures over time. *Topics in Language Disorders*, **25**, 243–253.
- Tyler, A. A., Lewis, K. E., Haskill, A. and Paul, K., 2003, Effects of a cycled morphological intervention on selected suppletive BE forms. *Clinical Linguistics and Phonetics*, **17**, 25–42.
- Tyler, A. A., Lewis, K. E., Haskill, A. and Tolbert, L. E., 2003, Outcomes of different speech and language goal attack strategies. *Journal of Speech, Language and Hearing Research*, **46**, 1077–1094.
- Tyler, A. A., Lewis, K. E. and Welch, C. M., 2003, Predictors of phonological change following intervention. *American Journal of Speech–Language Pathology*, **12**, 289–298.
- Tyler, A. A. and Sandoval, K. T., 1994, Preschoolers with phonological and language disorders: treating different linguistic domains. *Language, Speech, and Hearing Services in Schools*, **25**, 215–234.
- Tyler, A. A. and Watterson, K. H., 1991, Effects of phonological versus language intervention in preschoolers with both phonological and language impairment. *Child Language Teaching and Therapy*, **7**, 141–160.
- Tyler, A. A., Williams, M. J. and Lewis, K. E., 2006, Error consistency and the evaluation of treatment outcomes. *Clinical Linguistics and Phonetics*, **20**, 411–422.

6.3.4 Study Limitations

The systematic review had a specific remit to look at the evidence base related to intervention for SSD with preschool children (2yrs–5yrs 11mths). When we received reviews from our original submission there were some strongly voiced queries about why some papers were not included, leading to us constructing appendix C in the paper.

It was fortunate that we had had very strong and defensible inclusion and exclusion criteria from the outset. The biggest challenge to the inclusion of papers was the population of interest. Because we focused on ‘preschool’ children and required at least 80 percent to be within our defined age bracket and therefore some frequently cited papers were not included in the review.

One hundred and forty eight papers matched the inclusion criteria for the full Child Talk review. In order to ensure only high quality papers were included in the final stage, we followed previous researchers in using a score of ≥ 6 on the quality appraisal tools (PEDro-P and SCED) used to indicate a high-quality study (Maher, Sherrington, Herbert et al 2003; Camarinos & Marinko, 2009). A score of ≥ 6 was therefore used to determine the studies of acceptably high quality to be retained for inclusion in this work ($n = 58$).

An additional challenge for the review reported in paper 9 is that included papers had to contain an outcome measure which assessed speech output. The review did not include interventions that focused on prosodic skills or speech perception or other underlying speech processing skills unless these were included alongside a measurement of speech output. This was frequently reduced to mean length of utterance, which although clinically relevant, would have been better supported by a validated tool such as the Diagnostic Evaluation of Articulation and Phonology.

6.3.5 Contribution to knowledge and autoethnographic issues

6.3.5.1 Contribution to knowledge

This study systematically reviewed the evidence for those interventions that have been tested with children under 6 years of age. A model for classification of intervention studies in SSD is proposed and the evidence to support interventions within the model provided.

The evidence around interventions for children and service delivery can be complex. The paper provides a clear and transparent report about a facet (Speech) of practice. Speech and language therapists will be able to identify, at a glance, which interventions that have been tested with children under age 6 have evidence to support them. Evidence is varied in strength, and intervention studies using more robust research designs are needed to test fully the interventions described in the current literature.

Appendix 17 has an overview of how the research undertaken in paper 7 was disseminated prior to the paper's publication. It also provides a list of papers where paper 7 has been cited, and this is followed by a quality appraisal of the paper.

6.3.5.2 Autoethnographic issues

The majority of papers in this thesis have been written as team efforts. This was the first that required a discussion around 'Publication ethics.' The challenge with paper 9, was that I led the systematic review element of the Child Talk project. As such, paper 9 had originally been intended to be a Harding et al paper. However, we then included a model that was developed directly from the work of Yvonne Wren and her doctoral thesis. So who should be lead author?

Many journals publish a document outlining their ethical stance (Elsevier, 2017; Wiley, 2014), covering the duties of the publisher, editor, reviewers and authors. But, it seems that most people follow their previous experience as a guide to what they should do, and that this differs with disciplines. It was therefore interesting to discuss this with a range of people from different academic institutions. In some situations, and historically, authorship was given if you were in the team at any stage and no matter the level of input, in others you had to have made a significant contribution during the writing of the paper, but not necessarily at any other point.

Figure 5, lays out the criteria for authorship, and although from reading these it does feel unfair to those people who participate in the data collection, it is clear that significant involvement in analysis and paper preparation is required. What continues not to be specified is how to account for author order.

In preparing paper 9 for publication, the authors had a discussion which revolved around who the audience for the publication was, and who amongst us was most closely affiliated with that field of practice. This was clearly Yvonne Wren, and therefore she was identified as the corresponding author. In addition to the

development and use of her doctoral model we felt that she should be first author. This then led to me being second, as the person who had undertaken the systematic review, with Juliet Goldbart being third as the academic lead on this element of Child Talk and Sue Roulstone at fourth, as the CI on the project.

Although these discussions were more than amicable, I felt that it would have been good to have a justifiable, defensible set of regulations to follow. This would provide a way to take any emotion out of the decision if it had not been as easy to come to.

This has led me to read more around the topic and identify some tools published by the American Psychological Association, that I will use in the future (Winston, 1985; <https://www.apa.org/science/leadership/students/authorship-determination-scorecard.pdf>; <https://www.apa.org/science/leadership/students/authorship-tie-breaker-scorecard.pdf>). They also give me a clear set of principles to apply in discussion with people who seek my advice on this matter. As time has passed and my career has progressed, it has become clearer to me how important publications (and grants) are in progressing one's own career. It has also become clear to me that I am not very good at selling myself or fighting my corner in relation to this element of academic practice. Using these checklists to discuss the issue to authorship and author order objectively has and will continue to help me.

6.3.6 Future research questions

Child Talk's final report was published in 2015. Other than the publications written by the two PhD students that were part of the team (Blackwell, 2016; Davies, 2014), only one other paper had been published (Marshall, Harding & Roulstone, 2017) prior to paper 9. Since 2015, we have had a number of Child Talk articles that have been making slow progress in their production, but as no one in the team is now funded to work on the project or the dissemination it is hard to find the time. However, two more papers have been accepted for publication (Morgan, Marshall, Harding, et al 2019; Coad, Harding, Hambly, et al, 2020) and it is hoped that others will be ready for submission in the near future and that these will support the submission of grants to investigate parental understanding of speech and language disorders in preschool children.

Figure 5: Defining the Role of Authors

Criteria for authorship

The International Committee of Medical Journal Editors provides definitions of authors and contributors that are applicable in many instances beyond medical publishing. It recommends that authorship should be based on the following four criteria:

1. Substantial contributions to the conception or design of the work; or the acquisition, analysis, or interpretation of data for the work; AND
2. Drafting the work or revising it critically for important intellectual content; AND
3. Final approval of the version to be published; AND
4. Agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

All those designated as authors should meet all four criteria for authorship, and all who meet the four criteria should be identified as authors. Those who do not meet all four criteria should be acknowledged.

Authors collaborating on multidisciplinary and interdisciplinary work may have different and perhaps nonoverlapping areas of expertise. However, authors should still be able to stand "accountable" for ensuring investigation and resolution of "questions related to the accuracy or integrity of any part of the work."

By these criteria, acquisition of funding alone, collection of data alone, or general supervision of the research group alone does not constitute authorship. Also, each author should have participated sufficiently in the work to take public responsibility for appropriate portions of the content. This also applies to all authors designated within large multi-author groups and for on those occasions when authors report work on behalf of a larger group of investigators, upon which the International Committee of Medical Journal Editors provides guidance.

Journals should encourage authors of intercultural research to consider appropriate attribution for traditional knowledge, to the extent that this attribution does not compromise any agreed assurances of anonymity. This may include "traditional knowledge" notices, or citation of indigenous sources (such as people or community groups) or other cultural sources of knowledge by name within the text. In some fields, such as anthropology, appropriate attribution may require sharing authorship with intercultural collaborators and this may differ from the approach recommended by the International Committee of Medical Journal Editors. More information is at the [Australian Institute of Aboriginal and Torres Strait Islander Studies web site](#).

6.4 Summary

6.4.1 Summary of contribution to knowledge

All three papers had the aim of establishing current state of knowledge within the fields of investigation. In paper 7, I identified the factors previously used by other authors and groups used during the investigation of PPC in a HNC population.

Paper 8 was able to suggest that children with language delay may be less able to learn from their environment than their typically developing peers. Paper 9 produced a model for classification of interventions for SSD in preschool children, and provides researchers and practitioners with the current evidence.

6.4.2 Summary of autoethnographic issues

Systematic reviews are not intrinsically difficult, but they do require clear identification of the area of investigation, and following a process in rigorous fashion. Having someone experienced in the process, and with a demonstrable track record of success (publication), is seen as a benefit when working with teams that have limited experience. This is the role that I have been given by the Trust that employs me; I work with teams that are undertaking reviews which will be used to inform the development of future research bids. This frequently leads to being a co-applicant of these bids, and an increased portfolio of research work and experience.

Chapter 7 Discussion of Research Journey

The common research aim across the nine published papers that form this thesis has been to further our understanding of the positive, and negative, psychological change that can occur following a significant health intervention. All the participants in the reported studies have experienced a significant health related challenge and all have been in receipt of an evidence-based intervention to address this challenge.

Drawing this research together allows the reader to view the researcher's wider contribution to our understanding of psychological change. This thesis also allows me (the researcher) to reflect and more fully ground myself within my personal ethnography and discuss how this lens affects my past choices, and future plans.

7.1 Research apprenticeship

I completed my undergraduate degree in Psychology in 1998 and since then have worked with medical charities and the NHS, and across several medical specialities. In a way, I view this thesis as the summation of my 'research apprenticeship'. If I was to have planned what my apprenticeship was going to look like, I would not have been able to envisage the breadth and depth of experiences I have had the fortune to engage in.

There is no single pathway into conducting rigorous research. In my professional career, I support many aspiring researchers from across the NHS who approach research from a number of philosophical positions and work within a wide range of disciplines. A large part of my experience described within the papers presented here, has been to develop my own research expertise to deploy appropriately the correct methodological suite of tools to the research question at hand. Whilst my journey has been unique, there are some common themes for conducting research as a health psychologist.

If I were to plan a research apprenticeship, for someone else, I would provide extensive practical experience, under supervision, of all or most of the stages of an empirical research project, from the formulation of the research questions, through to research design and analysis, to the formal write-up of the research report.

In reflecting on my experiences and the skills I have acquired since my undergraduate degree and how I would plan helping someone else starting out in research, I would

set targets and outcomes to fall into three sets of skills: Research specific, discipline specific and personal.

7.1.1 Research methods skills

1. Explore extensively current research methodologies
2. Work closely and collaboratively with experienced researchers in the design and execution of research projects
3. Understand the appropriate research and analysis methods for each research question

7.1.2 Discipline - specific skills

4. Explore current research and theories in the specific field of health psychology under investigation
5. Evaluate critically the discipline and demonstrate a thorough working knowledge of the content of the research field
6. Consider ethical issues in research
7. Conduct high quality empirical research

7.1.3 Personal skills

8. Interact effectively and supportively within a research group
9. Communicate ideas, principles and theories effectively, fluently and professionally by written, graphic and oral means
10. Manage my own learning with minimum guidance using the full range of resources of the discipline
11. Seek and make use of feedback
12. Manage research as a project and know when to rule interesting things as out of scope but keep for a future project proposal
13. Engage effectively in debate and communicate effectively about research

This could also be restructured to mirror the Vitae Researcher Development Framework (<https://www.vitae.ac.uk/researchers-professional-development/about-the-vitae-researcher-development-framework>).

7.1.4 Range of setting and cross-professional working

In addition to the skills outlined above I have been able to work across multiple disease/disorder groups (head and neck cancer (HNC), Chronic Obstructive Pulmonary Disease (COPD), Speech and Language Therapy, etc.) with multi-disciplinary groups of professionals, within the charity sector (in units aligned to but not *in* the NHS), and the NHS, and also on multi-site projects. This has provided me with a range of experiences, and a variety of perspectives to use to inform and develop research undertaken with others and in formulating my own. I believe my exposure to a diverse range of settings has enabled me to become a unique researcher, and although I know it is more routine to specialise in a specific field of research, I would encourage anyone to stretch themselves before choosing their long term areas of specialism.

7.2 Contribution to new knowledge

Looking back across chapters 3, 4, 5 and 6, the work I have been part of and led has contributed to new knowledge across disciplines using a range of methodologies.

7.2.1 Contribution to knowledge of included single time point studies (Chapter 3)

Paper 1, presented the first study assessing the presence and prevalence of post-traumatic stress disorder (PTSD) in a COPD population. Whilst paper 2, built on a very limited number of publications investigating positive psychological change (PPC) following HNC. It found a relationship between biomedical, Health Related Quality of Life (HRQoL) and social factors in the development of PPC within a year of completion of treatment.

7.2.2 Contribution to knowledge of pre, post intervention studies (Chapter 4)

Paper 3 disseminated information on the Lung Information Needs Questionnaire (LINQ), the first tool designed with patients with COPD, to assess their information needs rather than their knowledge. The aim of this is to enable clinicians to identify areas requiring particular attention, during routine clinical practice.

Papers 4 and 5 build on the evidence base of psychosocial impact of Hyperbaric Oxygen Therapy on people treated for HNC, thereby allowing clinicians to include

these factors in their considerations as to whether to refer their patients for this treatment. Together these papers also contributed to our knowledge of how to design assessments that consider the patients' perspective across pre- and post-intervention.

7.2.3 Contribution to knowledge of longitudinal studies (Chapter 5)

Chapter 5 includes one paper (paper 6). The research presented within paper 6 created new knowledge through the identification and characterisation of trajectories in the development of PPC in a HNC population. Trajectories have previously been identified in a breast cancer population (Danhauer, 2015), but not with HNC. It furthered knowledge by covering a longer time span; over 5 years post cancer treatment. This represents the first truly longitudinal study within a cancer population, where previous research has stopped at 12 or 18 months following diagnosis or treatment.

Paper 6, further contributed to the knowledge base through the use of a cross-sequential methodology in a HNC population. The adoption of this cross-sequential methodology allowed for longitudinal data collection in a population that presents challenges for long term data collection due to the nature of the HNC and patterns of survival.

7.2.4 Contribution to knowledge of systematic reviews (Chapter 6)

All three papers in chapter 6 were systematic reviews aiming to establish current state of knowledge within their fields of investigation. In paper 7, I identified the factors previously used by other authors and research groups during the investigation of PPC in a HNC population. This informed my subsequent work, reported in papers 2 and 6.

Paper 8 was able to suggest that children with language delay may be less able to learn from their environment than their typically developing peers; whilst paper 9 produced a model for classification of interventions for Speech Sound Disorders (SSD) in preschool children.

Systematic reviews are a very well developed research tool and represent a complete domain in their own right. My work has not extended our knowledge of systematic reviews; however, their inclusion here is critical in fully describing the research arc of the other papers and my own understanding of the research process.

7.2.5 Summary of contribution to our psychological understanding of the impact of health interventions in relation to chronic conditions

This thesis presents nine publications for which I am the sole or joint author; sections 7.2.1 to 7.2.4 have given an overview of how the individual papers have contributed to knowledge of either the health condition under investigation, or the methodology used. Over the years since the research reported in the publications, as evidenced through the citations of the published articles (Appendices 1, 5, 7, 9, 12, 14, 18 & 19), it can be seen that this body of work has had a wider contribution to the field of enquiry.

The developed tools such as the LINQ (paper 3), and frameworks of practice (paper 9) have informed and influenced clinical practice, by providing clinicians with a tool that can be used in clinics to effectively understand a patients information needs (paper 3), or to select an evidence-based intervention for preschool children with a SSD. The papers have also shaped other researchers' activities (papers 1, 3, 7, 8, 9). They have informed the development of theoretical models (e.g. Furlong, Serry, Erickson & Morris, 2018), as well as understanding the impact of treatments in a specific cohort and generalising to a wider group of people. Examples of this are the use of papers 1 and 3 with people with COPD to inform work with cardiac patients' rehabilitation (Van Rotterdam, Hensley & Hazelton, 2019).

In addition to these points, the papers including Quality of Life (QoL) measures have become part of a sustained and growing movement within healthcare to define a positive outcome of treatment to be more than 'survival'. Where historically the Karnofsky Performance Scale Index was the sole measure requested to reflect a person's ability to perform activities of daily living (0=Dead – 100=Normal), it is now routine for databases to require at least a disease specific measure. Data disseminated as part of the included publications has allowed comparative analysis to be undertaken across groups of people with the same conditions and to be generalised to other health condition cohorts.

A further contribution to the wider field is methodological. The successful use of a cross-sequential design with a small incidence (16th most common) cancer cohort (HNC; paper 6) showed that this is a robust and appropriate methodology to use with these potentially hard to reach cohorts for longitudinal studies. This, combined with linear mixed-effects modelling, allows for incomplete multiple data returns to be

retained in the analysis, increasing the statistical power. This combination of study design and statistical analysis provided greater understanding and generalisability of factors effecting the development of the phenomenon under investigation (PPC in this instance).

7.3 Future direction of my research

In chapter 2, I outline the philosophical basis for my research. At the start of my research journey/apprenticeship I do not think I could have talked or written about my stance in formal terms. Whilst I may now have more research tools at my disposal, my underlying principles have not changed. I have always come from a realist perspective and used the pragmatics of that position to inform, but not dictate, the research methodology. I hope that I manage to stay involved in research that allows me to make findings that are directly relevant to patients and that can also be immediately (or at least rapidly) implemented into practice.

7.3.1 Changing face of research - Greater importance of PPI and making research directly relevant

An element of research that has developed over my time as a researcher is the importance of including the public in identifying important areas of research, formulating research questions, participating in, and helping with the analysis and dissemination of research, and all the bits in between. Historically, there was one tick box on the NHS ethics application to ask if Public Patient Involvement (PPI) had had any involvement in the development of a research project. Now there is an expectation that there *is* involvement and applicants are required to outline what it was, and how key stakeholders/PPI will continue to be involved. To evidence the strength of this issue, there is no way that you will be successful in obtaining NHS funding if you cannot demonstrate significant PPI involvement throughout the proposed research (<https://www.invo.org.uk/>).

The importance of PPI has always been central in my work, and it is wonderful to see it having an increasingly prominent position within medical research. Woven throughout the chapters of this thesis I have referred to how the people with conditions/illness have worked with me to inform both the development of the reported research and me. The people who have given their time, knowledge and

feedback have shown me the importance of their perspectives. The more I listen, the greater colour I can include in the interpretation of the data they provide.

The challenges that come with ensuring the voice of PPI is present from the conceptualisation to the promulgation of study are elements of work that I relish. My future research will continue to be strongly influenced and involve members of the public and patient cohorts.

7.4 Now bidding as a Chief Investigator

7.4.1 Novel health condition - Male Breast Cancer

Building on work I have been undertaking as a co-applicant and supported by both NIHR and research capability funding; I have developed a proposal to investigate the impact of a diagnosis of, and treatment for, male Breast Cancer (Appendix 21). The key moment that triggered this investigation came when I was in the Breast Care Centre waiting room prior to a meeting and a gentleman was called for his appointment. Although there were other men in the room, all conversation stopped and it felt as though everyone looked at him. It felt as if everyone had assumed that he was there to support his wife, not to be the patient. I raised this with the female clinician I was meeting with, and her response was, “Yes, I wonder what it is like for a man to have a women’s disease!”

Although the proposed work is small scale, I have already built a supportive multi-disciplinary team, with a plan for taking the work forward into a national study, for which we are currently seeking funding.

7.4.2 Novel methodologies – Drawing as a novel methodology for use with people experiencing HNC

While undertaking the research reported in papers 4 and 5, I had a lot of feedback from patients about the problems with interviews and questionnaires. Some of the problems were practical (poor eye sight, sore mouth and throat when talking), some were personal (don’t like questionnaires), all suggested that there must be other ways that they could let clinicians know about their experiences. I therefore undertook the work reported in the paper included in appendix 22 (Harding and Bradford, 2019). I have presented this work at international conferences, and following conversations with other delegates, was encouraged to seek funding to take it further. I am therefore currently in the process of working up a bid to build on

my previous work, and to include some of the team from the Head and Neck 5000 study (<http://www.headandneck5000.org.uk/>), and my colleagues at King's College London, Maxillofacial Prosthetics department.

7.4.3 Parent Child Interaction

Following on from the work undertaken as part of the Child Talk project (Roulstone et al, 2015), and reported in part in the systematic review papers 8 and 9, I worked with the team to develop a bid, which we sent to the Heather van der Lely Foundation Trust (<http://hvdfl.org.uk/>). The objective of the proposed work was to improve understanding of typical interactions in preschool children from a range of backgrounds in order to facilitate the future development of theory of Parent Child Interaction Therapy (PCIT) so that interventions are more appropriate and acceptable.

Although, we received good reviews, the grant was not funded. However, since then the trust has approached us directly and offered to fund a PhD to undertake the work proposed. I will be a supervisor on the PhD, and will be developing work that builds on other aspects of child talk, as well as the PhD work. Appendix 23 provides an overview of the PhD funded project.

The work I hope to develop directly from the PhD will compare or use the framework constructed with typically developing children, with a population of preschool children with Developmental Language Disorder. While the PhD work is being undertaken, I will seek funding to construct and validate a measure similar to the Illness Perception Questionnaire – Revised, for use with parents of children with Developmental Language Disorder in order that clinicians have a tool that will enable them to understand parental perspectives of the causes, timeline, consequences, and control of their child's condition.

7.5 Conclusion

The purpose of this thesis has been to present nine publications, for which I am the sole or joint author. These papers demonstrate my understanding of the research process and my ability to carry out research that meets the exacting standards required for peer-reviewed publications.

The span of included work contributes to our understanding of quality of life following having, and treatment for, a chronic health condition.

Paper 1 established the prevalence of post-traumatic stress disorder in a cohort of patients with chronic obstructive pulmonary disease patients. Paper 2 found that positive psychological change (as defined by the Silver Lining Questionnaire) was associated with, greater disease adversity overcome (survived), fewer disease and treatment side-effects, and higher health related quality of life. Both these papers were single-time-point studies.

Paper 3 described the development of a novel tool (Lung Information Needs Questionnaire) for understanding the information needs of patients referred for pulmonary rehabilitation. Paper 4 established a data-set of quality of life measures for patients undergoing hyperbaric oxygen therapy pre- and post- treatment for head and neck cancer. Paper 5 established the hyperbaric oxygen therapy impact on quality of life for patients suffering osteoradionecrosis following treatment for head and neck cancer. Papers 2, 4, and 5 were all pre- post- cohort studies.

Paper 6 identified and characterised a 5-year trajectory in the development of positive psychological change in a head and neck cancer population. Paper 6 also adopted a novel cross-sequential research methodology and linear mixed-effects model for data analysis.

Papers 7, 8, and 9 were systematic reviews establishing the current state knowledge for a domain and helping to identify research questions for those domains. Paper 7 examined extant measures for positive psychological change in head and neck cancer cohorts. Paper 8 highlighted the gap in understanding the relationship between parent and child language use during parent child interaction, and set the context for additional research in this area. Paper 9 presented a model for evaluating the evidence supporting different interventions for children with speech sound disorder.

The papers have been presented according to the methodology used. Surrounding the individual papers, I have given an overview of how I became involved in each project and then insights into the study limitations. I have then presented how each paper has contributed to knowledge and finally how being part of or leading the research has impacted on my development as an independent researcher.

References

- Achenbach, T.M. (1978). **Research in Developmental Psychology: Concepts, Strategies, Methods**. The Free Press, New York, NY.
- Adam, T.E., Holman Jones, S., Ellis, C. (2015). **Autoethnography Understanding Qualitative Research**. Oxford University Press: UK
- Ajzen, I. (1991). The theory of planned behavior. *Organizational Behavior and Human Decision Processes*. 50:179-211.
- Alonzo, A. A. (2000). The experience of chronic illness and post-traumatic stress disorder: the consequences of cumulative adversity. *Soc.Sci.Med.* 50:1475-1484.
- Aromataris, E., Fernandez, R., Godfrey, C., Holly, C., Kahlil, H., Tungpunkom, P. (2015). Summarizing systematic reviews: methodological development, conduct and reporting of an Umbrella review approach. *Int J Evid Based Health*. 13(3):132-40.
- Bauman-Waengler, J. (2000). *Articulation and phonological impairments: A clinical focus*. Needham Heights, MA: Allyn & Bacon.
- Bellizzi, K.M., Smith, A.W., Reeve, B.B., Alfano, C.M., Bernstein, L., Meeske, K., Baumgartner, K.B., Ballard-Barbash, R.R. (2010). Posttraumatic growth and health-related quality of life in a racially diverse cohort of breast cancer survivors. *J. Health Psychol.* 15:615–626. doi:10.1177/1359105309356364
- Berntsen, D., Rubin, D.C. (2006). The Centrality of Event Scale: A Measure of Integrating a Trauma into One's Identity and its Relation to Post-Traumatic Stress Disorder Symptoms. *Behav. Res. Ther.* 44:219–231. doi:10.1016/j.brat.2005.01.009
- Blackwell, A. (2016). Early language delay: a study of the evolving language environments of preschool children. <http://eprints.uwe.ac.uk/27407/>
- Booth, A., Fry-Smith, A. (2004). **Developing a research question**. In: Petticrew M, Roberts H, eds. *Systematic reviews in the social sciences*. Oxford: Blackwell.
- British Psychological Society. (2012). **Supplementary guidance on the use of Social Media**. <https://www.bps.org.uk/sites/bps.org.uk/files/Policy/Policy%20-%20Files/Supplementary%20Guidance%20on%20the%20Use%20of%20Social%20Media.pdf> (Accessed 26/08/2019)
- Broomfield, J., Dodd, B. (2004). Children with speech and language disability: caseload characteristics. *International Journal of Language and Communication Disorders*. 39 (3), 303-324.
- Brown, R. (1973) **First Language: the Early Stages**. Oxford, England: Harvard U. Press
- Calhoun, L.G., Tedeschi, R. (Eds.) (2006). **Handbook of Posttraumatic Growth: Research and Practice**. Routledge, Mahwah, N.J.
- Calhoun, L.G., Tedeschi, R. (1998). **Posttraumatic growth: future directions**. In: Tedeschi RG, Park CL, Calhoun LG, editors. *Posttraumatic growth: positive changes in the aftermath of crisis*. Mahwah, NJ: Lawrence Erlbaum Associates. pp. 215–238.
- Camarinos, J., Marinko, L. (2009) Effectiveness of manual physical therapy for painful shoulder conditions: a systematic review. *J Man Manip Ther.* 17:206-15. <http://dx.doi.org/10.1179/106698109791352076>

- Campbell, R.T. (1988). **Integrating conceptualization, design, and analysis in panel studies of the life course**. In: Methodological Issues in Aging Research. Springer-Verlag, New York, pp. 43–69.
- Cancer Research UK. (2015). **Oral cancer statistics** [WWW Document]. Cancer Res. UK. URL <http://www.cancerresearchuk.org/health-professional/cancer-statistics/statistics-by-cancer-type/oral-cancer> (Accessed 05.06.16).
- Chen, Z., Fan, V.S., Belza, B., Pike, K., Nguyen, H.Q. (2017). Association between Social Support and Self-Care Behaviors in Adults with Chronic Obstructive Pulmonary Disease. *Ann Am Thorac Soc*. 14(9):1419-1427. doi: 10.1513/AnnalsATS.201701-026OC.
- Chougule, P.B., Akhtar, M.S., Akerley, W., Ready, N., Safran, H., McRae, R., Nigri, P., Bellino, J., Koness, J., Radie-Keane, K., Wanebo, H. (1999). Chemoradiotherapy for advanced inoperable head and neck cancer: A phase II study. *Semin Radiat Oncol*. 9(2 Suppl 1):58-63.
- Coad, J., Harding, S., Hambly, H., Parker, N., Morgan, L., Marshall, J., Roulstone, S. (2020). Perspectives of Pre-school children in England with speech and language needs in the development of an evidence based framework of activities. *Child: Care, Health & Development* [In Press].
- Cook, C.E. (2008). Clinimetrics Corner: The Minimal Clinically Important Change Score (MCID): A Necessary Pretense. *The Journal of Manual & Manipulative Therapy*. 16(4): 82-83.
- Creswell, J.W. (1994). **Research Design: Qualitative and Quantitative Approaches**. Sage, Thousand Oaks.
- Critical Appraisal Skills Programme (2011). Critical Appraisal Skills Programme <https://casp-uk.net/> (Accessed 27/08/2019)
- Crosby, R.D., Kolotkin, R.L., Williams, G.R. (2003). Defining clinically meaningful change in health related quality of life. *J Clin Epidemiol*. 56:395–407.
- Danhauer, S.C., Crawford, S.L., Farmer, D.F., Avis, N.E. (2009). A longitudinal investigation of coping strategies and quality of life among younger women with breast cancer. *J. Behav. Med*. 32:371–379. doi:10.1007/s10865-009-9211-x
- Danhauer, S.C., Russell, G., Case, L.D., Sohl, S.J., Tedeschi, R.G., Addington, E.L., Triplett, K., Van Zee, K.J., Naftalis, E.Z., Levine, B., Avis, N.E. (2015). Trajectories of Posttraumatic Growth and Associated Characteristics in Women with Breast Cancer. *Ann. Behav. Med. Publ. Soc. Behav. Med*. 49:650–659. doi:10.1007/s12160-015-9696-1
- Davies, K. (2014). Parents' and speech and language therapists' roles in intervention for pre-school children with speech and language needs. <http://e-space.mmu.ac.uk/347077/>
- Dimopoulos, M.A., Chen, C., Spencer, A., Niesvizky, R., Attal, M., Stadtmauer, E.A., Petrucci, M.T., Yu, Z., Olesnyckyi, M., Zeldis, J.B., Knight, R.D., Weber, D.M. (2009). Long-term follow-up on overall survival from the MM-009 and MM-010 phase III trials of lenalidomide plus dexamethasone in patients with relapsed or refractory multiple myeloma. *Leukaemia*. 23(11), p.2147-2152.
- Dodd, B., Gillon, G. (2001). Exploring the relationship between phonological awareness, speech impairment, and literacy. *Advances in Speech-Language Pathology*, 3, 139-147.

Dorr, W., Hendry, J.H. (2001) Consequential late effects in normal tissues. *Radiother Oncol.* 61(3):223-31.

Elsevier (2017). **Ethical guideline for journal publication.**

https://www.elsevier.com/_data/assets/pdf_file/0009/300888/Ethical-guidelines-for-journal-publication-V2.0-May-2017-Elsevier.pdf (Accessed 26/08/19)

Farrington, D.P. (1991). Longitudinal research strategies: advantages, problems, and prospects. *J. Am. Acad. Child Adolesc. Psychiatry.* 30:369–374.
doi:10.1097/00004583-199105000-00003

Figueiredo, D., Cruz, J., Jácome, C., Marques, A. (2016). Exploring the Benefits to Caregivers of a Family-Oriented Pulmonary Rehabilitation Program. *Respir Care.* 61(8):1081-9. doi: 10.4187/respcare.04624. Epub 2016 Jul 5.

Foster, A.M., Worrall, L.E., Rose, M.L., O'Halloran, R. (2013). Turning the tide: putting acute aphasia management back on the agenda through evidence-based practice. *Aphasiology.* 27:420–43. <http://dx.doi.org/10.1080/02687038.2013.770818>

Frazier, P.A., Kaler, M.E. (2006). Assessing the validity of self-reported stress-related growth. *J. Consult. Clin. Psychol.* 74:859–869. doi:10.1037/0022-006X.74.5.859

Furlong, L., Serry, T., Erickson, S., Morris, M.E. (2018). Processes and challenges in clinical decision-making for children with speech-sound disorders. *Int J Lang Comm Disord.* 53(6). doi: 10.1111/1460-6984.12426

Gerlach, N.L., Barkhuysen, R., Kaanders, J.H., Janssens, G.O., Sterk, W., Merkx, M.A. (2008). The effect of hyperbaric oxygen therapy on quality of life in oral and oropharyngeal cancer patients treated with radiotherapy. *Int J Oral Maxillofac Surg.* 37(3):255-9. doi: 10.1016/j.ijom.2007.11.013. Epub 2008 Feb 11. PMID: 18262761

Goldstein, L.J., Gallagher, K.A., Bauer, S.M., Bauer, R.J., Baireddy, V., Liu, Z.J., Buerk, D.G., Thom, S.R., Velazquez, O.C. (2006). Endothelial progenitor cell release into circulation is triggered by hyperoxia-induced increases in bone marrow nitric oxide. *Stem Cells.* 24:2309-188

Grunwell, P. (1987). **Clinical phonology.** (2nd ed.). London: Croom Helm.

Gunty, A.L., Frazier, P.A., Tennen, H., Tomich, P., Tashiro, T., Crystal, P. (2011). Moderators of the relation between perceived and actual posttraumatic growth. *Psychological Trauma: Theory, Research, Practice and Policy.* 3(1):61-66.

Habermas, J. (1986). *Knowledge and Human Interests*, New Edition. Polity, Cambridge.

Habermas, J. (1991). *Communication and the Evolution of Society*, New Edition: Polity, Oxford.

Harding, S., Bradford, J. (2019). Drawing: A novel approach to understanding appearance change in people following treatment for head and neck cancer. *Sage Open Medicine.* 7:1-10

Harrington, S., McGurk, M., Llewellyn, C.D. (2008). Positive consequences of head and neck cancer: key correlates of finding benefit. *J. Psychosoc. Oncol.* 26:43–62.

Hassan, S.J., Weymuller, E.A. (1993). Assessment of quality of life in head and neck cancer patients. *Head Neck.* 15:485–496.

- Helgeson, V.S., Reynolds, K.A., Tomich, P.L. (2006). A meta-analytic review of benefit finding and growth. *J. Consult. Clin. Psychol.* 74:797–816. doi:10.1037/0022-006X.74.5.797
- Ho, S., Rajandram, R.K., Chan, N., Samman, N., McGrath, C., Zwahlen, R.A. (2011). The roles of hope and optimism on posttraumatic growth in oral cavity cancer patients. *Oral Oncol.* 47:121–124. doi:10.1016/j.oraloncology.2010.11.015
- Ho, S.M.Y., Chan, C.L.W., Ho, R.T.H. (2004). Posttraumatic growth in Chinese cancer survivors. *Psycho-oncology.* 13: 377-389.
- Holman, H., Lorig, K. (2000). Patients as partners in managing chronic disease. Partnership is a prerequisite for effective and efficient health Care. *BMJ.* 320 (7234):526-7
- Holtmaat, K., van der Spek, N., Cuijpers, P., Leemans, C.R., Verdonck-de Leeuw, I.M. (2016). Posttraumatic growth among head and neck cancer survivors with psychological distress. *Psycho-oncology.* 26(1):96-101. doi:10.1002/pon.4106
- Holman, H., Lorig, K. (2000). Patients as partners in managing chronic disease. Partnership is a prerequisite for effective and efficient health Care. *BMJ* 320 (7234). 526-7
- Howell, D., Molly, S., Wilkinson, K., Green, E., Orchard, K., Wang, K., Liberty, J. (2015). Patient reported outcomes in routine cancer clinical practice: a scoping review of use, impact on health outcomes, and implementation factors. *Annals of Oncology.* 26(9):1846-1858.
- Huffington Post. (2008). **The Huffington Post complete guide to blogging.** New York, NY: Simon & Schuster.
- Iglesias, C.P., Birks, Y.F., Torgerson, D.J. (2002). Increasing response rates to postal questionnaires. *BMJ.* 325, 444.
- Jaeschke, R., Singer, J., Guyatt, G. (1989). Measurement of health status. Ascertaining the minimal clinically important difference. *Controlled Clinical Trials.* 10:407-415.
- James, S. L. (1990). **Normal language acquisition.** Boston, MA: Allyn & Bacon
- McLeod, S. and Bleile, K. (2003). Neurological and developmental foundations of speech. Invited seminar presentation- American Speech-Language-Hearing Association Convention, Chicago.
- Janoff-Bulman, R., Frantz, C.M. (1997). The impact of trauma on meaning: From meaningless world to meaningful life. In: **The Transformation of Meaning in Psychological Therapies.** Wiley, New York, pp. 91–106.
- Janoff-Bulman, R., Schwartzberg, S. (1991). **Toward a general model of personal change. In: Handbook of Social and Clinical Psychology: The Health Perspective.** Pergamon Press, New York, pp. 488–508.
- Jenkinson, C., Chandola, T., Coulter, A., Bruster, S. (2001). An assessment of the construct validity of the SF-12 summary scores across ethnic groups. *J. Public Health Med.* 23:187–194.
- Jeong, J.H., Yoo, W.G. (2015). Effect of caregiver education on pulmonary rehabilitation, respiratory muscle strength and dyspnea in lung cancer patients. *J Phys Ther Sci.* 27(6):1653-4. doi: 10.1589/jpts.27.1653. Epub 2015 Jun 30.

- Joseph, S., Linley, A., Harris, G.J. (2004). Understanding Positive Change Following Trauma and Adversity: Structural Clarification. *J. Loss Trauma*. 10:83–96. doi:10.1080/15325020490890741
- Joseph, S., Linley, P.A. (Eds.) (2008). **Trauma, Recovery, and Growth: Positive Psychological Perspectives on Posttraumatic Stress**, 1 edition. John Wiley & Sons, Hoboken, N.J.
- Joseph, S., Linley, P.A. (2006). Growth following adversity: Theoretical perspectives and implications for clinical practice. *Clin. Psychol. Rev.* 26:1041–1053.
- Karajgi, B., Rifkin, A., Doddi, S., Kolli, R. (1990). The prevalence of anxiety disorders in patients with chronic obstructive pulmonary disease. *Am.J Psychiatry*. 147:200–201.
- Khafif, A., Posen, J., Yagil, Y., Beiser, M., Gil, Z., Ben-Yosef, R., Landsberg, R., Fliss, D.M. (2007). Quality of life in patients older than 75 years following major head and neck surgery. *Head Neck*. 29:932–939. doi:10.1002/hed.20635
- Kirk, J., Miller, M. (1991). **Reliability and Validity in Qualitative Research**. Sage Publications, New York, NY.
- Kraemer, H.C. (1994). Special methodological problems of childhood developmental follow-up studies: Focus on planning., in: **Developmental Follow-up: Concepts, Domains, and Methods**. Academic Press, San Diego, CA, pp. 259–76.
- Krosnick, J.A., Lavrakas, P.J., Kim, N. (2013). Survey Research. In 2nd ed. **Handbook of Research Methods in Social and Personality Psychology**. Cambridge University Press: Cambridge, UK.
- Kung, S., Rummans, T.A., Colligan, R.C., Clark, M.M., Sloan, J.A., Novotny, P.J., Huntington, J.L. (2006). Association of optimism-pessimism with quality of life in patients with head and neck and thyroid cancers. *Mayo Clin. Proc.* 81:1545–1552. doi:10.4065/81.12.1545
- Law, J., Boyle, J., Harris, F., Harkness, A., Nye C. (1998) Screening for speech and language delay: a systematic review of the literature. *Health Technology Assessment*, 2 (9).
- Law, J., Boyle, J., Harris, F., Harkness, A. Nye C. (2000) Prevalence and natural history of primary speech and language delay: findings from a systematic review of the literature. *International Journal of Language and Communication Disorders*. 35 (2) 165–188.
- Leong, A.M.F.I., Nik Jaafar, N.R., Zakaria, H., Rajandram, R.K., Mahadevan, R., Mohamad Yunus, M.R., Shah, S.A. (2015). Posttraumatic growth, depression and anxiety in head and neck cancer patients: examining their patterns and correlations in a prospective study. *Psycho-oncology*. 24:894–900. doi:10.1002/pon.3740
- Leventhal, H., Diefenbach, M., Leventhal, E.A. (1992). Illness cognition: Using common sense to understand treatment adherence and affect cognition interactions. *Cogn. Ther. Res.* 16:143–163. doi:10.1007/BF01173486
- Leventhal, H., Nerenz, D.R., Steele, D.F. (1984). Illness Representations and coping with health threats. In: **A Handbook of Psychology and Health**. Erlbaum, Hillside: NJ, pp. 219–252.
- Lim, L.L.-Y., Fisher, J.D. (1999). Use of the 12-item Short-Form (SF-12) Health Survey in an Australian heart and stroke population. *Qual. Life Res.* 8:1–8. doi:10.1023/A:1026409226544

- Linley, P.A., Joseph, S. (2004). Positive change following trauma and adversity: a review. *J. Trauma. Stress.* 17:11–21. doi:10.1023/B:JOTS.0000014671.27856.7e
- Llewellyn, C.D., Horney, D.J., McGurk, M., Weinman, J., Herold, J., Altman, K., Smith, H.E. (2013). Assessing the psychological predictors of benefit finding in patients with head and neck cancer. *Psycho-oncology.* 22:97–105. doi:10.1002/pon.2065
- Llewellyn, C.D., McGurk, M., Weinman, J. (2007). Illness and treatment beliefs in head and neck cancer: is Leventhal's common sense model a useful framework for determining changes in outcomes over time? *J. Psychosom. Res.* 63:17–26. doi:10.1016/j.jpsychores.2007.01.013
- Maher, C.G., Sherrington, C., Herbert, R.D., Moseley, A.M., Elkins, M. (2003). Reliability of the PEDro scale for rating quality of randomized controlled trials. *Phys Ther.* 83:713–21.
- Manne, S., Ostroff, J., Winkel, G., Goldstein, L., Fox, K., Grana, G. (2004). Posttraumatic Growth After Breast Cancer: Patient, Partner, and Couple Perspectives. *Psychosom. Med.* 66:442–454.
- Marshall, S., Haywood, K., Fitzpatrick, R. (2006). Impact of patient-reported outcome measures on routine practice: a structured review. *Journal of Evaluation in Clinical Practice.* 2006, 12(5):559-568.
- Marshall, J., Harding, S., Roulstone S. (2017). Language development, delay and intervention-the views of parents from communities that speech and language therapy managers in England consider to be under-served. *Int J Lang Commun Disord.* 52(4):489-500. doi: 10.1111/1460-6984.12288.
- Marx, R.E. (1999). Radiation injury to tissue. In Kindwall EP (ed). **Hyperbaric Medicine Practice.** 2nd Edition, Flagstaff, Best Publishing. Pp665-723
- McBride, O., Schroevers, M.J., Ranchor, A.V. (2009). The structure of adversarial growth in a sample of cancer patients 8 years post-diagnosis: a revision of the SLQ-38. *Psychol. Health.* 24:1197–1213. doi:10.1080/08870440802108900
- McBride, O.M.C., Dunwoody, L., Lowe-Strong, A., Kennedy, S.M. (2008). Examining adversarial growth in illness: the factor structure of the Silver Lining Questionnaire (SLQ-38). *Psychol. Health.* 23:661–678. doi:10.1080/14768320701356540
- McLeod, S., Bleile, K. (2003). Neurological and developmental foundations of speech. Invited seminar presentation- *American Speech-Language-Hearing Association* Convention, Chicago.
- McDowell, I. (2006). General Health Status and Quality of Life, in: **Measuring Health: A Guide to Rating Scales and Questionnaires.** Oxford University Press, Oxford, UK, pp. 520–702.
- McFarland, C., Alvaro, C. (2000). The impact of motivation on temporal comparisons: coping with traumatic events by perceiving personal growth. *J. Pers. Soc. Psychol.* 79:327–343.
- Melville, M.R., Lari, M.A., Brown, N., Young, T., Gray, D. (2003). Quality of life assessment using the short form 12 questionnaire is as reliable and sensitive as the short form 36 in distinguishing symptom severity in myocardial infarction survivors. *Heart Br. Card. Soc.* 89:1445–1446.
- Meredith, J.R., Raturi, A., Amoako-Gyampah, K., Kaplan, B. (1989). Alternative research paradigms in operations. *J. Oper. Manag.* 8:297–326.

- Miller, S.A. (2007). **Developmental Research Methods, Third Edition**. Sage Publications, Inc, Thousand Oaks, New York, NY.
- Miller, W.R., C'deBaca, J. (1994). Quantum change: Toward a psychology of transformation. In: **Can Personality Change?** American Psychological Association, Washington, D.C, pp. 253–280.
- Moola, S., Munn, Z., Tufanaru, C., Aromataris, E., Sears, K., Sfetcu, R., Currie, M., Qureshi, R., Mattis, P., Lisy, K., Mu, P-F. (2017). Chapter 7: Systematic reviews of etiology and risk. In: Aromataris E, Munn Z (Editors). **Joanna Briggs Institute Reviewer's Manual**. The Joanna Briggs Institute. Available from <https://reviewersmanual.joannabriggs.org/>
- Morgan, A.T., Vogel, A.P. (2009). Intervention for childhood apraxia of speech (Review). *Ur J Phys Rehabil Med*. 45(1):103-10.
- Morgan, L., Marshall, J., Harding, S., Powell, G., Wren, Y., Coad, J., Roulstone, S. (2019). 'It depends': Characterizing speech and language therapy for preschool children with developmental speech and language disorders. *Int J Lang Commun Disord*. 54(6):954-970. doi: 10.1111/1460-6984.12498.
- Munn, Z., Moola, S., Lisy, K., Riitano, D., Tufanaru, C. (2015). Methodological guidance for systematic reviews of observational epidemiological studies reporting prevalence and incidence data. *Int J Evid Based Health*. 13(3):147–153.
- National Collaborating Centre for Chronic Conditions. (2004). Chronic obstructive pulmonary disease. National clinical guideline on management of chronic obstructive pulmonary disease in adults in primary and secondary care. *Thorax*. 59 Suppl 1:1-232.
- National Institute for Clinical Excellence. (2004). **CG12 Chronic Obstructive Pulmonary Disease Clinical Guidelines**. London: Abba Litho Ltd.
- NHS Scotland. (2016). **Realist Medicine: Chief Medical Officer's Annual Report 2014-15**. (No. 0049/00492520). The Scottish Government, Edinburgh, Scotland.
- Office for National Statistics. (2000). **Mortality statistics: Cause, 1999**. (Rep. No. DH2 (No 26)). London: HMSO.
- Park, C.L., Cohen, L.H., Murch, R.L. (1996). Assessment and Prediction of Stress-Related Growth. *J. Pers*. 64:71–105. doi:10.1111/j.1467-6494.1996.tb00815.x
- Park, C.L., Helgeson, V.S. (2006). Introduction to the special section: growth following highly stressful life events – current status and future directions. *J Consult Clin Psychol*. 74(5):791-796.
- Parkes, C.M. (1975). What becomes of redundant world models? A contribution to the study of adaptation to change. *Br. J. Med. Psychol*. 48:131–137. doi:10.1111/j.2044-8341.1975.tb02315.x
- Pat-Horenczyk, R., Perry, S., Hamama-Raz, Y., Ziv, Y., Schramm-Yavin, S., Stemmer, S.M. (2015). Posttraumatic Growth in Breast Cancer Survivors: Constructive and Illusory Aspects. *J. Trauma. Stress*. 28:214–222. doi:10.1002/jts.22014
- Pawson, R., Tilley, N. (1997). **Evidence-Based Policy: A Realist Perspective**. Sage, London: UK.
- Pearlman, L.A., Saakvitne, K.W. (1995). **Trauma and the Therapist: Countertransference and Vicarious traumatization in psychotherapy with incest survivors**. New York: W.W. Norton

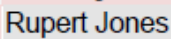
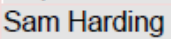
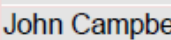
- Porter, S.R., Whitcomb, M.E. (2005). Non-response in student surveys: the role of demographics, engagement and personality. *Research in Higher Education*. 46:127-152.
- Rai, S.K., Yazdany, J., Fortin, P.R., Avina-Zubieta, J.A. (2015). Approaches for estimating minimal clinically important difference in systemic lupus erythematosus. *Arthritis Res Ther*. 17(1):143.
- Ransom, S., Sheldon, K.M., Jacobsen, P.B. (2008). Actual change and inaccurate recall contribute to posttraumatic growth following radiotherapy. *Journal of Consulting and Clinical Psychology*. 76(5):811-819.
- Rescorla, L., Schwartz, E. (1990). Outcome of toddlers with expressive language delay. *Applied Psycholinguistics*, 11, 393-407.
- Reed-Danahay, D. (2009). "Anthropologists, education and autoethnography". *Reviews in anthropology*. 38(1):32.
- Roberts, N.J., Kidd, L., Kirkwood, K., Cross, J., Partridge, M.R. (2018). A systematic review of the content and delivery of education in pulmonary rehabilitation programmes. *Respiratory Medicine*. 145:161-181.
- Rogers, S.N. (2001). Surgical principles and techniques for functional rehabilitation after oral cavity and oropharyngeal oncologic surgery. *Curr. Opin. Otolaryngol. Head Neck Surg*. 9:114–119.
- Rogers, S.N., D'Souza, J.J., Lowe, D., Kanatas, A. (2015). Longitudinal evaluation of health-related quality of life after osteoradionecrosis of the mandible. *Br. J. Oral Maxillofac. Surg*. 53:854–857. doi:10.1016/j.bjoms.2015.07.008
- Rogers, S.N., Gwanne, S., Lowe, D., Humphris, G., Yueh, B., Weymuller, E.A. (2002). The addition of mood and anxiety domains to the University of Washington quality of life scale. *Head Neck*. 24:521–529. doi:10.1002/hed.10106
- Rogers, S.N., Lowe, D., Brown, J.S., Vaughan, E.D. (1998). A comparison between the University of Washington Head and Neck Disease-Specific Measure and the Medical Short Form 36, EORTC QOQ-C33 and EORTC Head and Neck 35. *Oral Oncol*. 34: 361–372. doi:10.1016/S1368-8375(98)00031-1
- Rogers, S.N., Lowe, D., Brown, J.S., Vaughan, E.D. (2001). The relationship between length of stay and health-related quality of life in patients treated by primary surgery for oral and oropharyngeal cancer. *Int. J. Oral Maxillofac. Surg*. 30:209–215. doi:10.1054/ijom.2001.0040
- Rogers, S.N., Lowe, D., Humphris, G. (2000). Distinct patient groups in oral cancer: a prospective study of perceived health status following primary surgery. *Oral Oncol*. 36:529–538.
- Roulstone, S., Marshall, J., Powell, G., Goldbart, J., Wren, Y., Coad, J., Daykin, N., Powell, J.E., Lascelles, L., Hollingworth, W., Emond, A., Peters, T.J., Pollock, J.I., Fernandes, C., Moultrie, J., Harding, S.A., Morgan, L., Hambly, H.F., Parker, N.K., Coad, R.A. (2015). Evidence-based intervention for preschool children with primary speech and language impairments: Child Talk - an exploratory mixed-methods study. *Programme Grants for Applied Research*. 3(5).
<https://www.journalslibrary.nihr.ac.uk/programmes/pgfar/RP-PG-0109-10073/#/>
- Rubin, P., Cassarett, G.W. (1968). **Clinical Radiation Pathology**. Philadelphia: W.B. Saunders Company

- Sackett, D.L., Rosenberg, W.M., Gray, J.A., Haynes, R.B., Richardson, W.S. (1996). Evidence based medicine: what it is and what it isn't. *BMJ*. 312:71–2. <http://dx.doi.org/10.1136/bmj.312.7023.71>
- Salsman, J.M., Segerstrom, S.C., Brechting, E.H., Carlson, C.R., Andrykowski, M.A. (2009). Posttraumatic growth and PTSD symptomatology among colorectal cancer survivors: a 3-month longitudinal examination of cognitive processing. *Psychooncology*. 18: 30–41. doi:10.1002/pon.1367
- Schoen, P.J., Reintsema, H., Raghoobar, G.M., Vissink, A., Roodenburg, J.L. (2004). The use of implant retained mandibular prostheses in the oral rehabilitation of head and neck cancer patients. A review and rationale for treatment planning. *Oral Oncol*. 40(9):862-71.
- Sears, F.W., Zemansky, M.W., Young, H.D. (1987). **University Physics**. 7th ed. Addison-Wesley Publishing Company, Reading, Massachusetts.
- Sears, S.R., Stanton, A.L., Danoff-Burg, S. (2003). The yellow brick road and the emerald city: benefit finding, positive reappraisal coping and posttraumatic growth in women with early-stage breast cancer. *Health Psychol. Off. J. Div. Health Psychol. Am. Psychol. Assoc.* 22: 487–497. doi:10.1037/0278-6133.22.5.487
- Sheikh, A.I., Marotta, S.A. (2005). A cross-validation study of the Posttraumatic Growth Inventory. *Measurement and Evaluation in Counselling and Development*. 38: 66-77.
- Shriberg, L. D. (2010). Childhood Speech Sound Disorders: From postbehaviourism to the post genomic era. In Paul, R. and Flipsen Jr, P. (Eds) *Speech sound disorders in children*. San Diego, California; Plural Publishing.
- Silva, S.M., Crespo, C., Canavarro, M.C. (2012). Pathways for psychological adjustment in breast cancer: a longitudinal study on coping strategies and posttraumatic growth. *Psychol. Health*. 27:1323–1341. doi:10.1080/08870446.2012.676644
- Sivo, S.A., Saunders, C., Chang, Q., Jiang, J.J. (2006). How Low Should You Go? Low Response Rates and the Validity of Inference in IS Questionnaire Research. *J. Assoc. Inf. Syst.* 7(6). <https://aisel.aisnet.org/jais/vol7/iss6/17>
- Sloan, L. (2016) **The SAGE handbook of social media research methods**. Sage publications
- Sodergren, S.C., Hyland, M.E. (1998). Qualitative phase in the development of the Silver Lining Questionnaire. *Qual. Life Res.* 6, 365.
- Sodergren, S.C., Hyland, M.E. (2000). What are the positive consequences of illness? *Psychol. Health*. 15:85–97. doi:10.1080/08870440008400290
- Sodergren, S.C., Hyland, M.E., Crawford, A., Partridge, M.R. (2004). Positivity in illness: self-delusion or existential growth? *Br. J. Health Psychol.* 9:163–174. doi:10.1348/135910704773891023
- Sodergren, S.C., Hyland, M.E., Singh, S.J., Sewell, L. (2002). The Effect of Rehabilitation on Positive Interpretations of Illness. *Psychol. Health*. 17:753–760. doi:10.1080/0887044021000009674
- Steel, J.L., Gamblin, T.C., Carr, B.I. (2008). Measuring post-traumatic growth in people diagnosed with hepatobiliary cancer: directions for future research. *Oncol. Nurs. Forum*. 35:643–650. doi:10.1188/08.ONF.643-650

- Stoll, C., Schelling, G., Goetz, A.E., Kilger, E., Bayer, A., Kapfhammer, H.P., Rothenhäusler, H.B., Kreuzer, E., Reichart, B., Peter, K. (2000). Health-related quality of life and post-traumatic stress disorder in patients after cardiac surgery and intensive care treatment. *J. Thorac. Cardiovasc. Surg.* 120:505–512.
- Sumalla, E.C., Ochoa, C., Blanco, I. (2009). Posttraumatic growth in cancer: reality or illusion? *Clin. Psychol. Rev.* 29:24–33. doi:10.1016/j.cpr.2008.09.006
- Tedeschi, L.G.C., Richard G. (1996). **Trauma & Transformation: Growing in the Aftermath of Suffering**. Sage Publications, Incorporated, Thousand Oaks, Calif.
- Tedeschi, R.G., Calhoun, L.G. (2004). Posttraumatic Growth: Conceptual Foundations and Empirical Evidence. *Psychol. Inq.* 15: 1–18. doi:10.1207/s15327965pli1501_01
- Tedstone, J. E., Tarrier, N. (2003). Posttraumatic stress disorder following medical illness and treatment. *Clin Psychol Rev.* 23:409–448.
- Tennen, H., Affleck, G. (2002). Benefit-finding and benefit-reminding, in: **The Handbook of Positive Psychology**. Oxford University Press, New York, pp. 584–594.
- Terrell, J.E., Nanavati, K.A., Esclamado, R.M., Bishop, J.K., Bradford, C.R., Wolf, G.T. (1997). Head and neck cancer-specific quality of life: instrument validation. *Arch. Otolaryngol. Head Neck Surg.* 123:1125–1132.
- The Cochrane Collaboration. (1999). **Guidance on the conduct of narrative synthesis in systematic reviews: a comparison of guidance-led narrative synthesis versus meta-analysis**. <https://abstracts.cochrane.org/2006-dublin/guidance-conduct-narrative-synthesis-systematic-reviews-comparison-guidance-led> (Accessed 27/08/2019)
- Thornton, A.A. (2002). Perceiving benefits in the cancer experience. *J. Clin. Psychol. Med. Settings.* 9:153–165.
- Tinati, R., Halford, S., Carr, L., Pope, C. (2014). Big data: methodological challenges and approaches for sociological analysis. *Sociology.* 48:663–81.
- Trott, K.R. (1984). Chronic damage after radiation therapy: Challenge to radiation biology. *Int J Radat Oncol Biol Phys.* 10:907–913.
- Updegraff, J.A., Taylor, S.E. (2000). From vulnerability to growth: Positive and negative effects of stressful life events. In: **Loss and Trauma: General and Close Relationship Perspectives**. Brunner-Routledge, Philadelphia, P.A., pp. 3–28.
- Valderas, J.M., Kotzeva, A., Espallargues, M., Guyatt, G., Ferrans, C.E., Halyard, M.Y., Revicki, D.A., Symonds, T., Parada, A, Alonso, J. (2008). The impact of measuring patient-reported outcomes in clinical practice: a systematic review of the literature. *Qual Life Res.* 17(2):179–193.
- Van Rotterdam, J., Hensley, M. Hazelton, M. (2019). A comparative effectiveness review: responsiveness of patient outcome measure in cardiac and pulmonary rehabilitation, *Journal of Cardiopulmonary rehabilitation and prevention.* 39(2):73–84.
- Wagena, E. J., Arrindell, W. A., Wouters, E. F., van Schayck, C. P. (2005). Are patients with COPD psychologically distressed? *Eur Respir J.* 26:242–248.
- Wang, A.W.T., Chang, C.S., Chen, S.T., Chen, D.R., Hsu, W.Y. (2014). Identification of posttraumatic growth trajectories in the first year after breast cancer surgery. *Psycho-oncology.* 23:1399–1405. doi:10.1002/pon.3577

- Ware, J., Kosinski, M., Keller, S.D. (1996). A 12-Item Short-Form Health Survey: construction of scales and preliminary tests of reliability and validity. *Med. Care*. 34:220–233.
- Ware, J., Kosinski, M., Keller, S.D. (2001). **How to score the SF-12 Physical and Mental Health Summary Scale**. Health Institute, New England Medical Center, Boston.
- Weiss, T., Berger, R. (2006). Reliability and validity of a Spanish version of the Posttraumatic Growth Inventory. *Research on Social Work and Practice*. 16:191-199.
- Wells, G., Beaton, D., Shea, B., Boers, M., Simon, L., Strand, V., Brooks, P., Tugwell, P. (2001). Minimal clinically important differences: review of methods. *J Rheumatol*. 28:406-412.
- White, C.A. 2001. **Cognitive Behaviour Therapy for Chronic Medical Problems. A Guide to Assessment and Treatment in Practice**. Chichester: Wiley.
- White, R., Walker, P., Roberts, S., Kalisky, S., White, P. (2006). Bristol COPD Knowledge Questionnaire (BCKQ): testing what we teach patients about COPD. *Chronic Respiratory Disease*. 3:123-131.
- Wiley. (2014). **Best Practice Guideline on Publishing Ethics: A publisher's perspective** 2nd Edition. <https://authorservices.wiley.com/asset/Best-Practice-Guidelines-on-Publishing-Ethics-2ed.pdf> (Accessed 26/08/19)
- Winston, Jr., R. B. (1985). A suggested procedure for determining order of authorship in research publications. *Journal of Counseling and Development*. 63:515-518.
- Wohlwill, J.F. (1973). **The Study of Behavioral Development**. Academic Press, New York, NY.
- Wolchik, S.A., Coxe, S., Tein, J.Y., Sandler, I.N., Ayers, T.S. (2008). Six-Year Longitudinal Predictors of Posttraumatic Growth in Parentally Bereaved Adolescents and Young Adults. *Omega J. Death Dying*. 58:107–128. doi:10.2190/OM.58.2.b
- Wren, Y. (2005). An evaluation of the use of computers in phonology therapy. PhD Thesis. University of Bristol.
<https://ethos.bl.uk/OrderDetails.do?did=1&uin=uk.bl.ethos.422606>
- Wren, Y., Miller, L. L., Peters, T. J., Emond, A., & Roulstone, S. (2016). Prevalence and predictors of persistent speech sound disorder at eight years old: Findings from a population cohort study. *Journal of Speech, Language, and Hearing Research*, 59, 647–673. doi:10.1044/2015_JSLHR-S-14-0282
- Yalom, I.D., Lieberman, M.A. (1991). Bereavement and heightened existential awareness. *Psychiatry*. 54:334–345.
- Zoellner, T., Maercker, A. (2006). Posttraumatic growth in clinical psychology - a critical review and introduction of a two component model. *Clin. Psychol. Rev*. 26:626–653. doi:10.1016/j.cpr.2006.01.008

Appendix 1: Contribution to publication forms

Research and Knowledge Exchange Graduate School Form RDPUB (ROUTE 1 AND 2)		 Manchester Metropolitan University
PhD BY PUBLISHED WORK (ROUTE 1/2): CONTRIBUTION TO PUBLICATIONS		
<i>This form is to accompany an application for registration for PhD where the PhD is by Published Work. A separate form should be completed for <u>each</u> publication that is submitted with the proposal and should accompany the RD1 form.</i>		
1. The Candidate		
First Name(s):	Sam	Preferred Title: Dr
Surname:	Harding	
MMU e-mail address:	samantha.harding2@stu.mmu.ac.u	Contact Number: 07944 363973
Personal e-mail address:	sharding.jb@gmail.com	Student ID Number: 18050275
2. Title of PhD Proposal		
Psychological understanding of the impact of health interventions in relation to chronic conditions.		
3. Title of Research Output		
The prevalence of posttraumatic stress disorder in patients undergoing pulmonary rehabilitation and changes in PTSD symptoms following rehabilitation.		
4. Candidate's contribution to the research output (State nature and approximate percentage contribution of each author)		
The candidate was involved in the concept, design, and majority in the data collection and analysis and paper writing. Jones RC 30%, Harding SA 30% Chung MC 15%, Campbell J 25%		
5. Co author(s):		
I confirm that the contribution indicated above is an accurate assessment of the contribution by the candidate to the research output named in section 3.		
Name	Signature	Current e-mail address
Rupert Jones	 Rupert Jones	rupert.jones@plymouth.ac.uk
Sam Harding	 Sam Harding	sharding.jb@gmail.com
John Campbell	 John Campbell	john.campbell@exeter.ac.uk
6. Statement by Director of Studies/Advisor		
I confirm that I have read the above publication and am satisfied that the extent and nature of the candidate's contribution is as indicated in section 4 above.		
Signature: 	Date:	
(Director of Studies/Advisor)		
7. Signature of Faculty Research Degrees Administrator		
Signature: 	Date:	
(Faculty Research Degrees Administrator)		

**Research and Knowledge Exchange
Graduate School
Form RDPUB (ROUTE 1 AND 2)**



**PhD BY PUBLISHED WORK (ROUTE 1/2):
CONTRIBUTION TO PUBLICATIONS**

This form is to accompany an application for registration for PhD where the PhD is by Published Work. A separate form should be completed for each publication that is submitted with the proposal and should accompany the RD1 form.

1. The Candidate

First Name(s):	Sam	Preferred Title:	Dr
Surname:	Harding		
MMU e-mail address:	samantha.harding2@stu.mmu.ac.uk	Contact Number:	07944 363973
Personal e-mail address:	sharding.jb@gmail.com	Student ID Number:	18050275

2. Title of PhD Proposal

Psychological understanding of the impact of health interventions in relation to chronic conditions.

3. Title of Research Output

The impact of treatment for head and neck cancer on positive psychological change within a year of completing treatment.

**4. Candidate's contribution to the research output
(State nature and approximate percentage contribution of each author)**

95% - SH wrote the manuscript
5% - TM commented on the manuscript and suggested revisions

5. Co author(s):

I confirm that the contribution indicated above is an accurate assessment of the contribution by the candidate to the research output named in section 3.

Name	Signature	Current e-mail address
Sam Harding	Sam Harding Digitally signed by Sam Harding Date: 2018.03.03 16:27:57 Z	sharding.jb@gmail.com
Tim Moss	Tim Moss Digitally signed by Tim Moss Date: 2018.03.08 15:48:59 Z	Tim.Moss@uwe.ac.uk

6. Statement by Director of Studies/Advisor

I confirm that I have read the above publication and am satisfied that the extent and nature of the candidate's contribution is as indicated in section 4 above.

Signature: Date:

(Director of Studies/Advisor)

7. Signature of Faculty Research Degrees Administrator

Signature: Date:

(Faculty Research Degrees Administrator)

Research and Knowledge Exchange
Graduate School
Form RDPUB (ROUTE 1 AND 2)



**PhD BY PUBLISHED WORK (ROUTE 1/2):
CONTRIBUTION TO PUBLICATIONS**

This form is to accompany an application for registration for PhD where the PhD is by Published Work. A separate form should be completed for each publication that is submitted with the proposal and should accompany the RD1 form.

1. The Candidate

First Name(s):	Sam	Preferred Title: Dr
Surname:	Harding	
MMU e-mail address:	samantha.harding2@stb.smm.ac.uk	Contact Number: 07944 363973
Personal e-mail address:	sharding.jb@gmail.com	Student ID Number: 18050275

2. Title of PhD Proposal

Psychological understanding of the impact of health interventions in relation to chronic conditions

3. Title of Research Output

Educational Impact of pulmonary rehabilitation: Lung Information Needs Questionnaire.

4. Candidate's contribution to the research output

(State nature and approximate percentage contribution of each author)

The candidate was involved in the concept, design, ethics approval and conduct of the study including data collection and to a limited degree in the analysis, she also made important contributions to the manuscript. Jones RC 25% Wang X 30%, Harding S 25%, Bolt J 5% Hyland M 15%

5. Co author(s):

I confirm that the contribution indicated above is an accurate assessment of the contribution by the candidate to the research output named in section 3.

Name	Signature	Current e-mail address
Rupert Jones	<i>Rupert Jones</i>	Digitally signed by Rupert Jones Date: 2018.03.06 17:47:09 Z rupert.jones@plymouth.ac.uk
Sam Harding	<i>Sam Harding</i>	Digitally signed by Sam Harding Date: 2018.03.07 14:59:34 Z sharding.jb@gmail.com
Michael Hyland	<i>M Hyland</i>	M.Hyland@plymouth.ac.uk

6. Statement by Director of Studies/Advisor

I confirm that I have read the above publication and am satisfied that the extent and nature of the candidate's contribution is as indicated in section 4 above.

Signature: _____ Date: _____
(Director of Studies/Advisor)

7. Signature of Faculty Research Degrees Administrator

Signature: _____ Date: _____
(Faculty Research Degrees Administrator)

Research and Knowledge Exchange
Graduate School
Form RDPUB (ROUTE 1 AND 2)



**PhD BY PUBLISHED WORK (ROUTE 1/2):
CONTRIBUTION TO PUBLICATIONS**

This form is to accompany an application for registration for PhD where the PhD is by Published Work. A separate form should be completed for each publication that is submitted with the proposal and should accompany the RD1 form.

1. The Candidate

First Name(s):	Sam	Preferred Title:	Dr
Surname:	Harding		
MMU e-mail address:	samantha.harding2@stu.mmu.ac.uk	Contact Number:	07944 363973
Personal e-mail address:	sharding.jb@gmail.com	Student ID Number:	187050275

2. Title of PhD Proposal

Psychological understanding of the impact of health interventions in relation to chronic conditions.

3. Title of Research Output

Impact of perioperative hyperbaric oxygen therapy on the quality of life of maxillofacial patients who undergo surgery in irradiated fields.

4. Candidate's contribution to the research output

(State nature and approximate percentage contribution of each author)

85% - SH undertook the research and wrote the manuscript. The other three authors all commented on the drafts of the manuscript and approved submission of the paper.

5. Co author(s):

I confirm that the contribution indicated above is an accurate assessment of the contribution by the candidate to the research output named in section 3.

Name	Signature	Current e-mail address
Sam Harding	Sam Harding Digitally signed by Sam Harding Date: 2016.03.03 17:27:05 Z	sharding.jb@gmail.com
Simon Hodder	Simon Hodder Digitally signed by Simon Hodder DN: cn=Simon Hodder, o=MMU, email=Simon.Hodder@wales.nhs.uk, c=GB Date: 2016.03.03 11:30:57	Simon.Hodder@wales.nhs.uk
David Courtney	Mr David Courtney Digitally signed by Mr David Courtney Date: 2016.03.12 16:58:10 Z	david.courtney@nhs.net

6. Statement by Director of Studies/Advisor

I confirm that I have read the above publication and am satisfied that the extent and nature of the candidate's contribution is as indicated in section 4 above.

Signature: Date: _____
(Director of Studies/Advisor)

7. Signature of Faculty Research Degrees Administrator

Signature: Date: _____
(Faculty Research Degrees Administrator)

Research and Knowledge Exchange
Graduate School
Form RDPUB (ROUTE 1 AND 2)



**PhD BY PUBLISHED WORK (ROUTE 1/2):
CONTRIBUTION TO PUBLICATIONS**

This form is to accompany an application for registration for PhD where the PhD is by Published Work. A separate form should be completed for each publication that is submitted with the proposal and should accompany the RD1 form.

1. The Candidate

First Name(s):	Sam	Preferred Title:	Dr
Surname:	Harding		
MMU e-mail address:	samantha.harding2@stu.mmu.ac.u	Contact Number:	07944 363973
Personal e-mail address:	sharding.jb@gmail.com	Student ID Number:	18050275

2. Title of PhD Proposal

Psychological understanding of the impact of health interventions in relation to chronic conditions.

3. Title of Research Output

Effects of hyperbaric oxygen therapy on quality of life in maxillofacial patients with type III osteoradionecrosis.

4. Candidate's contribution to the research output

(State nature and approximate percentage contribution of each author)

85% - SH undertook the research and wrote the manuscript. The other three authors all commented on the drafts of the manuscript and approved submission of the paper.

5. Co author(s):

I confirm that the contribution indicated above is an accurate assessment of the contribution by the candidate to the research output named in section 3.

Name	Signature	Current e-mail address
Sam Harding	Sam Harding Digitally signed by Sam Harding Date: 2018.03.03 17:27:05 Z	sharding.jb@gmail.com
David Courtney	Mr David Courtney Digitally signed by Mr David Courtney Date: 2018.03.03 21:58:03 Z	david.courtney@nhs.net
Simon Hodder	Simon Hodder Digitally signed by Simon Hodder DN: cn=Simon Hodder, o=MMU, ou=Faculty of Health, email=Simon.Hodder@wales.nhs.uk, c=GB Date: 2018.03.03 17:50:02 Z	Simon.Hodder@wales.nhs.uk

6. Statement by Director of Studies/Advisor

I confirm that I have read the above publication and am satisfied that the extent and nature of the candidate's contribution is as indicated in section 4 above.

Signature: Date: _____
(Director of Studies/Advisor)

7. Signature of Faculty Research Degrees Administrator

Signature: Date: _____
(Faculty Research Degrees Administrator)

Research and Knowledge Exchange
Graduate School
Form RDPUB (ROUTE 1 AND 2)



**PhD BY PUBLISHED WORK (ROUTE 1/2):
CONTRIBUTION TO PUBLICATIONS**

This form is to accompany an application for registration for PhD where the PhD is by Published Work. A separate form should be completed for each publication that is submitted with the proposal and should accompany the RD1 form.

1. The Candidate

First Name(s):	Sam	Preferred Title:	Dr
Surname:	Harding		
MMU e-mail address:	samantha.harding2@stu.mmu.ac.uk	Contact Number:	07944363973
Personal e-mail address:	sharding.jb@gmail.com	Student ID Number:	18050275

2. Title of PhD Proposal

Psychological understanding of the impact of health interventions in relation to chronic conditions.

3. Title of Research Output

The trajectory of positive psychological change in a head and neck cancer population.

4. Candidate's contribution to the research output

(State nature and approximate percentage contribution of each author)

100% - Sole author of the article.

5. Co author(s):

I confirm that the contribution indicated above is an accurate assessment of the contribution by the candidate to the research output named in section 3.

Name	Signature	Current e-mail address
Sam Harding	Sam Harding Digitally signed by Sam Harding Date: 2018.03.03 16:18:48 Z	sharding.jb@gmail.com

6. Statement by Director of Studies/Advisor

I confirm that I have read the above publication and am satisfied that the extent and nature of the candidate's contribution is as indicated in section 4 above.

Signature: Date:

(Director of Studies/Advisor)

7. Signature of Faculty Research Degrees Administrator

Signature: Date:

(Faculty Research Degrees Administrator)

**Research and Knowledge Exchange
Graduate School
Form RDPUB (ROUTE 1 AND 2)**



**PhD BY PUBLISHED WORK (ROUTE 1/2):
CONTRIBUTION TO PUBLICATIONS**

This form is to accompany an application for registration for PhD where the PhD is by Published Work. A separate form should be completed for each publication that is submitted with the proposal and should accompany the RD1 form.

1. The Candidate

First Name(s):	Sam	Preferred Title:	Dr
Surname:	Harding		
MMU e-mail address:	samantha.harding2@stu.mmu.ac.uk	Contact Number:	07944 363973
Personal e-mail address:	sharding.jb@gmail.com	Student ID Number:	18050275

2. Title of PhD Proposal

Psychological understanding of the impact of health interventions in relation to chronic conditions.

3. Title of Research Output

Existence of benefit finding and posttraumatic growth in people treated for head and neck cancer: a systematic review.

4. Candidate's contribution to the research output

(State nature and approximate percentage contribution of each author)

85% - SH - Lead the majority of the work to produce the systematic review including the writing of the manuscript
10% - FS - Check the inclusion of papers, quality checked the papers and proofed the manuscript
5% - TM - Reviewed and revised the manuscript

5. Co author(s):

I confirm that the contribution indicated above is an accurate assessment of the contribution by the candidate to the research output named in section 3.

Name	Signature	Current e-mail address
Sam Harding	Sam Harding Digitally signed by Sam Harding Date: 2016.03.03 16:41:01 Z	sharding.jb@gmail.com
Fatemeh Sani Pour	Fatemeh Sani Pour Digitally signed by Fatemeh Sani Pour DN: cn=Fatemeh Sani Pour, o=MMU, email=sani.pour@mmu.ac.uk, c=GB Date: 2016.03.03 17:40:22 Z	F.SaniPour@salford.ac.uk
Tim Moss	Tim Moss Digitally signed by Tim Moss Date: 2016.03.06 15:50:20 Z	Tim.Moss@uwe.ac.uk

6. Statement by Director of Studies/Advisor

I confirm that I have read the above publication and am satisfied that the extent and nature of the candidate's contribution is as indicated in section 4 above.

Signature: Date: _____
(Director of Studies/Advisor)

7. Signature of Faculty Research Degrees Administrator

Signature: Date: _____
(Faculty Research Degrees Administrator)

**Research and Knowledge Exchange
Graduate School
Form RDPUB (ROUTE 1 AND 2)**



**PhD BY PUBLISHED WORK (ROUTE 1/2):
CONTRIBUTION TO PUBLICATIONS**

This form is to accompany an application for registration for PhD where the PhD is by Published Work. A separate form should be completed for each publication that is submitted with the proposal and should accompany the RD1 form.

1. The Candidate

First Name(s):	Sam	Preferred Title:	Dr
Surname:	Harding		
MMU e-mail address:	samantha.harding2@stu.mmu.ac.u	Contact Number:	07944 363973
Personal e-mail address:	sharding.jb@gmail.com	Student ID Number:	18050275

2. Title of PhD Proposal

Psychological understanding of the impact of health interventions in relation to chronic conditions.

3. Title of Research Output

Characteristics of parent child interactions: A systematic review of studies comparing children with primary language impairment and their typically developing peers.

4. Candidate's contribution to the research output

(State nature and approximate percentage contribution of each author)

All refined and agreed review question. AB (70%) identified titles, excluded based on abstract/full text, extracted data, conducted narrative synthesis and wrote review. 10% abstract check by SH, 10% full text check by SB and SR, included consensus discussion. All reviewed and agreed manuscript.

5. Co author(s):

I confirm that the contribution indicated above is an accurate assessment of the contribution by the candidate to the research output named in section 3.

Name	Signature	Current e-mail address
Anna Blackwell	Anna Blackwell Digitally signed by Anna Blackwell Date: 2018.04.05 16:02:54 +01'00'	anna.blackwell@bristol.ac.uk
Selma Babayigit	Selma babayigit Digitally signed by Selma babayigit Date: 2018.11.12 17:10:37 Z	Selma.Babayigit@uwe.ac.uk
Sue Roulstone	Susan Roulstone Digitally signed by Susan Roulstone Date: 2018.05.17 22:38:29 +01'00'	Susan.Roulstone@uwe.ac.uk

6. Statement by Director of Studies/Advisor

I confirm that I have read the above publication and am satisfied that the extent and nature of the candidate's contribution is as indicated in section 4 above.

Signature:  Date: _____
(Director of Studies/Advisor)

7. Signature of Faculty Research Degrees Administrator

Signature:  Date: _____
(Faculty Research Degrees Administrator)

**Research and Knowledge Exchange
Graduate School
Form RDPUB (ROUTE 1 AND 2)**



**PhD BY PUBLISHED WORK (ROUTE 1/2):
CONTRIBUTION TO PUBLICATIONS**

This form is to accompany an application for registration for PhD where the PhD is by Published Work. A separate form should be completed for each publication that is submitted with the proposal and should accompany the RD1 form.

1. The Candidate

First Name(s):	Sam	Preferred Title:	Dr
Surname:	Harding		
MMU e-mail address:	samantha.harding2@stu.mmu.ac.uk	Contact Number:	07944 363973
Personal e-mail address:	sharding.jb@gmail.com	Student ID Number:	18050275

2. Title of PhD Proposal

Psychological understanding of the impact of health interventions in relation to chronic conditions.

3. Title of Research Output

A systematic review and classification of interventions for speech-sound disorder in preschool children.

4. Candidate's contribution to the research output

(State nature and approximate percentage contribution of each author)

Sam led the work on this paper through carrying out the systematic review and critical appraisal of the included papers. She also wrote the methods and results section of the paper. Her contribution to this paper was 60% with co-authors Wren completing 30% and Goldbart and Roulstone 5% each.

5. Co author(s):

I confirm that the contribution indicated above is an accurate assessment of the contribution by the candidate to the research output named in section 3.

Name	Signature	Current e-mail address
Yvonne Wren	 Digitally signed by Yvonne Wren Date: 2018.03.04 15:54:19 Z	yvonne.wren@bristol.ac.uk
Juliet Goldbart	 Digitally signed by Juliet Goldbart Date: 2018.03.05 09:42:54 Z	J.Goldbart@mmu.ac.uk
Sue Roulstone	 Digitally signed by Susan Roulstone Date: 2018.04.04 15:08:04 +0100	susan.roulstone@uwe.ac.uk

6. Statement by Director of Studies/Advisor

I confirm that I have read the above publication and am satisfied that the extent and nature of the candidate's contribution is as indicated in section 4 above.

Signature: Date: _____
(Director of Studies/Advisor)

7. Signature of Faculty Research Degrees Administrator

Signature: Date: _____
(Faculty Research Degrees Administrator)

Appendix 2: Impact of Research – Paper 1

Prior to the publication of the paper two poster presentations were given. One was at the Society for Academic Primary Care, a national conference in Keele, UK (Appendix 3), and the other was at the American Thorax Society annual conference in San Diego, California.

The Journal of Cardiopulmonary Rehabilitation and Prevention at the time of writing has an impact factor of 1.685, in 2009 it was 1.550. It also had a Source Normalized Impact per Paper value of 0.701. It is currently ranked 86/128 in cardiac and cardiovascular system journals. It is the official journal of the American Association of Cardiovascular and Pulmonary Rehabilitation and the Canadian Association of Cardiovascular Prevention and Rehabilitation. This journal was chosen not only because of its IF, but because of the audience. The journal is sent to all the members of two associations, thus having a direct impact on their patients care and as such could directly change their practice after being exposed to this paper. It was also selected because of its indexing (Journal of Cardiopulmonary Rehabilitation and Prevention is cited in Allied and Complementary Medicine Database, Cumulative Index to Nursing Administration and Health Literature, EBSCO A-Z, EMBASE, Ex Libris, HINARI, JournalGuide, MEDLINE, ProQuest, PubMed, Science Citation Index Expanded, Scopus, TDNet, and Web of Science.), meaning that it is readily findable on literature searches.

Subsequent to the publication of the paper Mendeley indicate that it has been cited **12** times, 8 of which can be identified through Web of Science services:

- 1) Chung MC, Jones RCM, Harding SA, Campbell J (2016) Posttraumatic Stress Disorder among Older Patients with Chronic Obstructive Pulmonary Disease. *Psychiatric Quarterly*. 87(4):605-618.
- 2) Harrison SL, Lee A, Janaudis-Ferreira T, Goldstein RS, Brooks D (2016) Mindfulness in people with a respiratory diagnosis: A systematic review. *Patient Educ Couns*. 99(3): 348-355. doi: 10.1016/j.pec.2015.10.013.
- 3) Zimmermann Teieria PJ, Porto L, Kristensen CH, et al (2015) Post-traumatic Stress Symptoms and Exacerbations in COPD Patients. *Journal of Chronic Obstructive Pulmonary Disease*. 12(1):90-95.

- 4) Abrams TE, Blevins A, Weg MW (2015) Chronic obstructive lung disease and posttraumatic stress disorder: current perspectives. *Int J Chron Obstruct Pulmon Dis.* 10:2219-33. doi: 10.2147/COPD.S71449.
- 5) Disler RT, Currow DC, Phillips JL et al (2012) Interventions to support a palliative care approach in patients with chronic obstructive pulmonary disease: An integrative review. *International Journal of Nursing Studies.* 49(11): 1443-1458.
- 6) Nair HP, Ekenga CC, Cone JE, Brackbill RM, Farfel MR, Stellman SD (2012) Co-occurring lower respiratory symptoms and posttraumatic stress disorder 5 to 6 years after the World Trade Center terrorist attack. *Am J Public Health.* 102(10):1964-73. doi: 10.2105/AJPH.2012.300690.
- 7) Baumschlager D, Haas-Krammer A, Rothenhaeusler HB (2011) Emotional status, cognitive performance and quality of life in HIV-Infected patients. *Nervenarzt.* 8(7): 902-909.
- 8) De Miranda S, Pochard F, Chaize M, et al (2011) Postintensive care unit psychological burden in patients with chronic obstructive pulmonary disease and informal caregivers: a multi-center study. *Critical Care Medicine.* 39(1): 112-118

Other indices of impact are also available. For example Mendeley Stats give the author a measure of readership. It indicates how many people registered with Scopus, who have linked their accounts with the paper, and how want to follow the use of the paper in other publications. The current paper has a Mendeley statistic of 36. The challenge with this statistic is that as a standalone article we cannot interpret how impactful this is.

The quality of the paper can be judged rigorously using a standardised Quality Appraisal (QA) tools. Each paper included in the thesis has been quality appraised by myself and an independent clinical researcher (Phil Clatworthy; <https://www.nbt.nhs.uk/our-services/a-z-consultants/dr-philip-clatworthy>). We used the Joanna Briggs Institutes range of tools and selected the one which most appropriately fitted the methodology of the research. As with systematic review process, differences in our ratings were discussed and consensus reached.

Quality Appraisal of paper 1 - Prevalence Data (Munn, Moola, Lisy et al 2015)

	Yes	No	Unclear	Not applicable
1. Was the sample frame appropriate to address the target population?	☒	☐	☐	☐
2. Were study participants sampled in an appropriate way?	☒	☐	☐	☐
3. Was the sample size adequate?	☐	☒	☐	☐
4. Were the study subjects and the setting described in detail?	☒	☐	☐	☐
5. Was the data analysis conducted with sufficient coverage of the identified sample?	☒	☐	☐	☐
6. Were valid methods used for the identification of the condition?	☒	☐	☐	☐
7. Was the condition measured in a standard, reliable way for all participants?	☒	☐	☐	☐
8. Was there appropriate statistical analysis?	☒	☐	☐	☐
9. Was the response rate adequate, and if not, was the low response rate managed appropriately?	☒	☐	☐	☐

Assigning a score of 2 for 'Yes', 1 for 'Unclear' and 0 for 'No' and 'Not applicable', this paper receives a total of 16/18 or 89%. As mentioned above (Section 3.1.4.1) there are significant limitations with the sample of participants.

Appendix 3: Paper 1 poster presentation



The prevalence of posttraumatic stress disorder in patients with chronic obstructive pulmonary disease undergoing pulmonary rehabilitation.

Rupert Jones¹, Sam Harding¹, Man Cheung Chung², John Campbell¹.
¹Primary Care, Peninsula Medical School, ²University of Plymouth

Research questions:

1. What is the prevalence of PTSD in patients with COPD undergoing pulmonary rehabilitation (PR)?
2. Do PTSD symptom scores falls after PR?

Background

PTSD is a common, serious condition, which is often undetected. It is treatable.¹

Main features: hyperarousal, avoidance behaviour and re-experiencing after a traumatic experience.²

Little is known about the prevalence in chronic diseases such as COPD and the impact of PTSD on the symptoms and disability.

Methods:

Participants: Patients with COPD referred to PR programmes in Plymouth, Exeter and East Devon.

Outcome measures:

- The Posttraumatic Diagnostic Scale
- Revised Impact of Events scale
- SF 12

Normal PR outcomes also recorded:

- The incremental shuttle walking test
- Hospital Anxiety and Depression score (HADS)
- Chronic Respiratory Questionnaire (CRQ)

References: 1. NICE guidelines on PTSD, 2005.
 2. DSM-4 American Psychiatric Association, 1995.




Results:

Participants
 100 subjects took part, mean age 68yrs (8.2); 65 (65%) male; 17 (17%) current smokers; mean 45 pack years. 70 (70%) completed rehabilitation.

PTSD
 The prevalence of PTSD was 8%; Those with PTSD had worse health status: mean CRQ total scores: 76 v 60; $p=0.02$ mean HADS anxiety scores: 11.8 v 7.2; $p=0.01$

Rehabilitation
 Completion of PR was associated with improved exercise capacity, quality of life (all CRQ scales and anxiety and depression) significantly in this cohort of 100 patients. However there was no change in PTSD symptom severity.

Conclusion

PTSD was present in a small minority of COPD patients referred to PR and is associated with poor health status.

There is no evidence that PTSD symptoms fall after rehabilitation, despite its positive effects on HAD scores, exercise and health status.

Dr. Jones is supported by a DoH Researcher Development Award



PENINSULA
 MEDICAL SCHOOL
UNIVERSITIES OF EXETER & PLYMOUTH

Appendix 4: Description of questionnaires used in paper 2

Both a QoL and a HRQoL measure were included in the current work as they investigate different levels of the QoL concept. Quality of Life is a broad concept which covers all aspects of life. The SF-12 is a health status QoL measure. The authors of the SF-12 aimed to develop a short, generic measure of subjective health status that was psychometrically sound and could be applied to a wide range of settings (Ware *et al.*, 1996). The UoW was included as a HRQoL measure. It focuses on the effects of illness and specifically on the impact treatment may have on QoL. Quality of Life (in this study the SF-12) is therefore broader than HRQoL (UoW) because it includes evaluation of related, non-disease specific features of life whereas HRQoL is connected to an individual's disease status. HRQoL can help understand the distinction between aspects of life related to health.

Medical Outcomes Short Form 12 Version 2

The Medical Outcomes Short Form 12 Version 2 (SF-12) is a multipurpose, short-form questionnaire with only 12 items, all selected from the SF-36 Health Survey (Ware *et al.*, 1996). It is a generic measure, as opposed to one that targets a specific age, disease, or treatment group. It has been proven to be useful in measuring outcomes in clinical trials. The SF-12 was selected for use in this study as it is well validated but brief enough to minimise response burden (Melville *et al.*, 2003; Stoll *et al.*, 2000).

The SF-12 is a self-administered survey designed to measure health concepts across age, disease and treatment groups. It takes approximately 3 minutes to complete, which is a significant advantage compared with the SF-36, which takes 5 to 10 minutes per respondent (Melville *et al.*, 2003). It tests eight health domains: 4 of physical health (physical functioning, role functioning, bodily pain and general health perception), and 4 of mental health (social functioning, role mental, vitality and mental health).

Results for each participant are expressed in terms of two meta-scores; a Physical Component Summary (PCS) and a Mental Component Summary (MCS). To calculate the PCS and MCS, test items are scored and normalised in an algorithm described in the SF-12 users' manual (Ware *et al.*, 2001). Raw data are transformed into norm-based scores for each domain (population mean 50, standard deviation 10) and PCS and MCS measures for each participant are calculated using factor score coefficients derived from the general population. This process yields PCS and MCS scores with a

range of 0 to 100. Scores greater than 50 represent above average health status, whereas people with a score of 40 function at a level lower than 84% of the population (one standard deviation), and people with a score less than 30 function at a level lower than approximately 98% of the population (two standard deviations).

Psychometric status of the SF-12 has been found adequate by studies determining its concurrent validity with the SF-36. Using data from 10 general American population surveys ($n = 24,293$) the SF-12 achieved multiple R squares of 0.91 and 0.92 in predictions of the SF-36 PCS and MCS respectively (Ware, *et al.*, 1996). The SF-12 has demonstrated internal consistencies of $\alpha = 0.71 - 0.90$ for all of the sub-scales including the PCS and MCS and test-retest of between $r = 0.71$ and 0.84 (Ware, *et al.*, 1996).

This measure has been used in many fields of health care research including HNC. Khafif *et al.* (2007) investigated generic HRQoL with this measure and its association with QoL specifically related to HNC. Other researchers have utilised the SF-12 to compliment the investigate of optimism and pessimism, disfigurement and dysfunction and how people with HNC represent their illness in relation to Leventhal's common sense model (Kung *et al.*, 2006; Llewellyn *et al.*, 2007; Terrell *et al.*, 1997).

Silver Lining Questionnaire

The 38-item Silver Lining Questionnaire (SLQ; Sodergren and Hyland, 2000) was selected for its psychometric properties and its reported ability to measure the extent to which people believe their illness has had a positive benefit despite the negative consequences of being ill.

The SLQ measures 10 facets of positivity with illness. These comprise: restructuring of life; reappraisal of life; spiritual gains; self-improvement; self-awareness; skills and new pursuits; sensitivity to emotions; confrontation of current concerns; improved interpersonal relationships and positive consequences for others. People respond using five categories: strongly agree, agree, not sure, disagree, and strongly disagree. An overall score is obtained by scoring each item as 1 for responses strongly agree and agree, and 0 for all the other response options. The total score therefore reflects the total number of items with which the respondent agrees (i.e. the number of positive consequences of illness experienced), and varies between 0 (low positivity, i.e. the respondent agrees with no items) and 38 (high positivity, i.e. the respondent

agrees with all items). Cronbach's alpha is 0.93 and retest reliability is $r = 0.90$ (Sodergren *et al.*, 2002).

The authors of the SLQ refer to it measuring adversarial growth. Although it has not been used to investigate PPC in people specifically with or following HNC, it has been used with mixed cancer cohorts (breast, colorectal, gynaecological, and lung) and found that adversarial growth is related to cancer stage (McBride *et al.*, 2009). When assessed in the setting of a range of diseases including a mixed cancer cohort, the severity of the illness affects the level of PPC reported using the SLQ (Sodergren *et al.*, 2004; Sodergren and Hyland, 2000).

No data is currently available on how long it takes to complete the SLQ.

University of Washington quality of life questionnaire

This is a broad measure of HRQoL specifically designed for use with people who have or have had HNC. It has good patient acceptability, practicality, validity, reliability and responsiveness (Hassan and Weymuller, 1993). Not only does the University of Washington (UoW) measure HRQoL items, it can also predict outcomes such as length of stay in hospital (Rogers *et al.*, 2001) and identify discrete patient groups (Rogers *et al.*, 2000). The UoW (Hassan and Weymuller, 1993) quality of life (QoL) questionnaire version 4 was used and covers 12 domains: pain, appearance, activity, recreation, swallowing, chewing, speech, shoulder function, taste, saliva, mood and anxiety. Each question is scaled from 0 (worst) to 100 (best) according to the hierarchy of response. There are also three global questions, one asking about HRQoL compared to the month before they had cancer, one asking about HRQoL during the past 7 days and the third asking about overall QoL during the past 7 days.

The UoW has been validated by comparison to the Karnofsky Scale and Sickness Impact Profile demonstrating an average criterion validity of 0.85 (Hassan and Weymuller, 1993). It has also been found to have internal consistency between $\alpha = 0.80$ and 0.79 and Test-retest of $r = 0.91$ (Rogers *et al.*, 2002).

The questionnaire was designed to be a self-completed measure specific to HNC patients. Hassan and Weymuller (1993) suggest that the advantages of the UoW are that it is brief and self-administered and, from the clinician's perspective, it is multi-factorial, allowing sufficient detail to identify subtle change as well as providing questions specific to HNC. It allows no input from the health provider, thus reflecting QoL as indicated by the patient.

Appendix 5: Impact of research of paper 2

As with paper one, I undertook some lower level dissemination activities. The first of these was a poster presentation given at the British Psychological Society BPS Annual meeting in London in 2012 (Appendix 6). I then presented this work at the 9th International Conference Head Neck Cancer Quality of Life. The audience for this conference was a mixture of medics, nurses and allied health professionals, most speech and language therapists. The third presentation was given to a national meeting of the Royal College of Speech and Language Therapist HNC specialist interest group. The International Journal of Oral and Maxillofacial Surgery at the time of writing this thesis has an impact factor of 2.164 and a Source Normalized Impact per Paper of 1.553. It is currently ranked 24/200 in journals of dentistry, oral surgery and medicine and 77/200 of surgical journals. International Journal of Oral and Maxillofacial Surgery is the journal of the International Association of Oral and Maxillofacial Surgeon. As such International Journal of Oral and Maxillofacial Surgery is available free to International Association of Oral and Maxillofacial Surgeon members. Importantly International Journal of Oral and Maxillofacial Surgery is indexed on some of the key databases (Current Contents / Clinical Medicine, EMBASE/Excerpta Medica, Medline/Index Medicus, Medical Documentation Service, Research Alert, ISI Science Citation Index, SciSearch, BIOSIS/Biological Abstracts, Scopus), ensuring it can be found by medics and other scientists, nurses, allied health professionals and researchers.

The paper was published in 2018 and as such has only been cited once, and that citation was in my subsequent longitudinal paper:

- 1) Harding SA (2018) The trajectory of positive psychological change in a head and neck cancer population. International Journal of Oral and Maxillofacial Surgery. 47(5):578-584

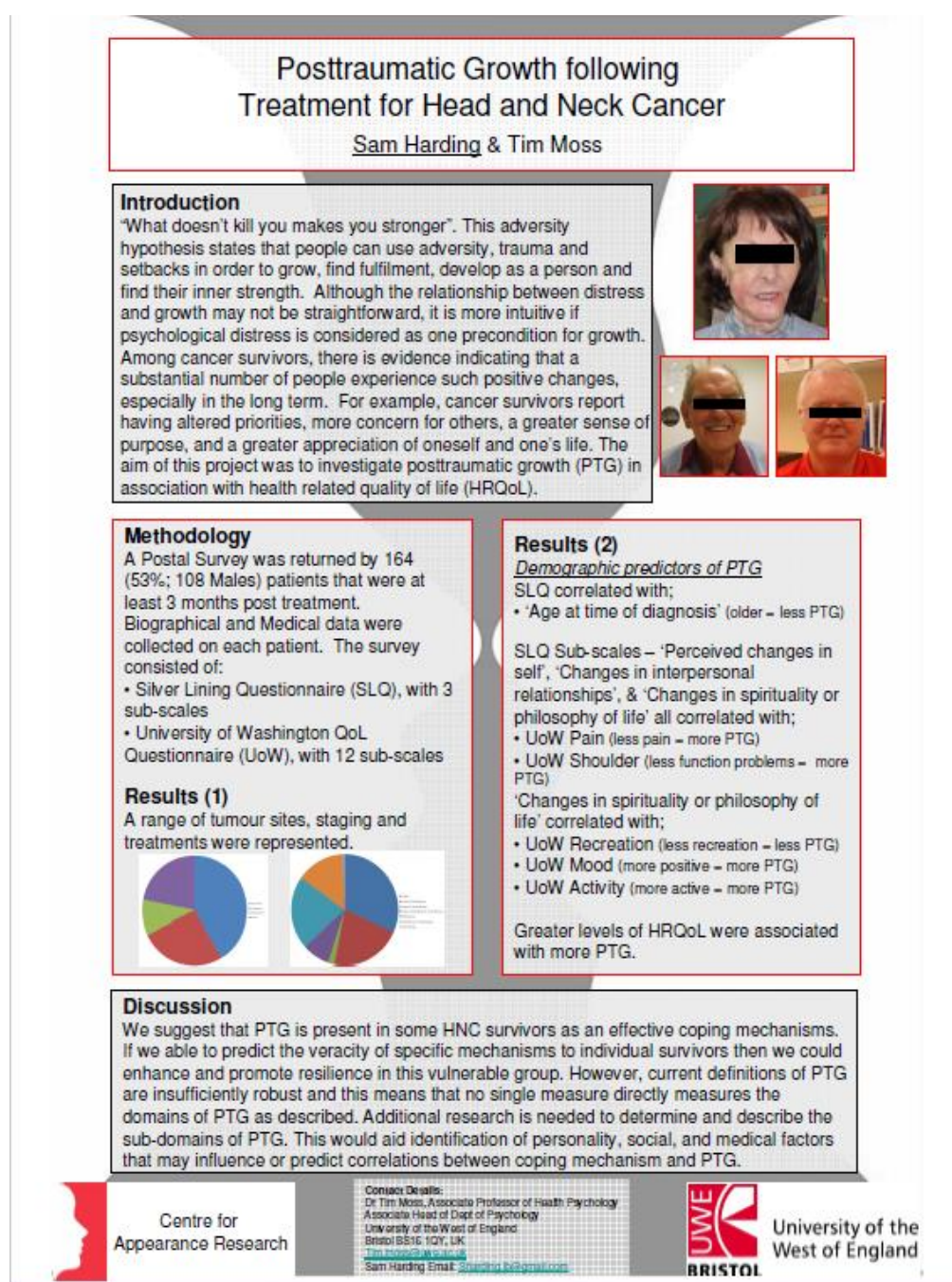
Mendeley Stats for paper 2 are at the time of writing 13. In comparison to 36 of the paper, it would appear that it is of less interest to researchers' and clinicians. However, this paper has been in circulation for approximately 18 months, as opposed to the 9 years of paper 1. It is therefore encouraging that this paper in a niche field may be stimulating interest.

As with paper 1 Dr Phil Clatworthy and I applied a QA tool. Once again we used the Joanna Briggs Institute's range of tools and for paper 2 we used the Case Series checklist.

Quality Appraisal - Case Series (Moola, Munn, Tufanaru et al, 2017)

	Yes	No	Unclear	Not applicable
1. Were there clear criteria for inclusion in the case series?	☒	☐	☐	☐
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	☒	☐	☐	☐
3. Were valid methods used for identification of the condition for all participants included in the case series?	☒	☐	☐	☐
4. Did the case series have consecutive inclusion of participants?	☐	☒	☐	☐
5. Did the case series have complete inclusion of participants?	☐	☒	☐	☐
6. Was there clear reporting of the demographics of the participants in the study?	☒	☐	☐	☐
7. Was there clear reporting of clinical information of the participants?	☒	☐	☐	☐
8. Were the outcomes or follow up results of cases clearly reported?	☐	☐	☐	☒
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	☐	☒	☐	☐
10. Was statistical analysis appropriate?	☒	☐	☐	☐

Appendix 6: Paper 2 Poster presentation



Appendix 7: Impact of research of paper 3

The team started to disseminate work around the development and usefulness of the LINQ in 2004. Nine poster presentations were given at national and international conferences such as European Respiratory Society , Thorax and European General Practice Research Network (Appendix 8, and I gave two oral presentations were given at the European Health Psychology Conference and the South West Society for Academic Primary Care. The paper was published in Respiratory Medicine, which at the time of writing has an impact factor of 3.230 and a Source Normalized Impact per Paper value of 1.17. It is an international journal with a focus on publishing clinically relevant research. Unlike the Journal of Cardiopulmonary Rehabilitation and Prevention, this is not a journal of a respiratory organisation. So, it does not provide the automatic dissemination that that type of journal brings. However, it is indexed in the main databases (Scopus, AIDS Abstracts, SIIC Data Bases, Current Contents/Life Sciences and Clinical Medicine, MEDLINE®, Excerpta Medica, Science Citation Index, Current Awareness in Biological Sciences, EMBASE), and as such can readily identified as evidenced by paper 3's inclusion in a systematic review (Roberts, Kidd, Kirkwood et al, 2018).

At the time of writing, Mendeley indicates that it has been cited **21** times, all of which can be identified through Web of Science services:

- 1) Roberts NJ, Kidd L, Kirkwood K, Cross J, Partridge MR. (2018) A systematic review of the content and delivery of education in pulmonary rehabilitation programmes. Respiratory Medicine. 145: 161-181
- 2) Ward S, Sewell L, Singh S. (2018) Evaluation of multidisciplinary pulmonary rehabilitation education delivered by either DVD or spoken talk. Clinical Respiratory Journal. 12(11): 2546-2550
- 3) Mayer AF, Gulart AA, Dos Santos K, (...), Karoh M, O'Neill B. (2018) Translation, cross-cultural adaptation, and reliability of the understanding COPD questionnaire for use in Brazil | [Tradução, adaptação transcultural e confiabilidade do questionário Understanding COPD para uso no Brasil]. Jornal Brasileiro de Pneumologia 44(4): 285-291
- 4) Blackstock FC, Lareau SC, Nici L, (...), Meek PM, Singh S. (2018) Chronic obstructive pulmonary disease education in pulmonary rehabilitation: An official American thoracic society/thoracic society of Australia and New

Zealand/Canadian thoracic society/British thoracic society workshop report.
Annals of the American Thoracic Society. 15(7): 769-784

5) Guilleminault L, Rolland Y, Didier A. (2018) Characteristics of non-pharmacological interventions in the elderly with COPD. Smoking cessation, pulmonary rehabilitation, nutritional management and patient education | [Particularités de la prise en charge non médicamenteuse de la BPCO chez les sujets âgés. Réhabilitation, sevrage tabagique, nutrition et éducation thérapeutique]. Revue des Maladies Respiratoires 35(6): 626-641

6) Clari M, Ivziku D, Casciaro R, Matarese M. (2018) The Unmet Needs of People with Chronic Obstructive Pulmonary Disease: A Systematic Review of Qualitative Findings. COPD: Journal of Chronic Obstructive Pulmonary Disease. 15(1): 79-88

7) Early F, Young JS, Robinshaw E, (...), Mi EZ, Fuld JP. (2017) A case series of an off-the-shelf online health resource with integrated nurse coaching to support self-management in COPD. International Journal of COPD. 12, pp. 2955-2967

8) Sandelowsky H, Krakau I, Modin S, Stallberg B, Nagar A. (2017) Case Method in COPD education for primary care physicians: Study protocol for a cluster randomised controlled trial. Trials 18(1):197

9) Rosero-Carvajal HE, Cuero-Campaz DF, Arias-Balanta, Á.J., Wilches-Luna, E.C. (2017) Pilot study. Changes in the score of the Lung Information Needs Questionnaire (LINQ) after an educational intervention in patients with chronic respiratory disease | [Estudio piloto. Mudanças na pontuação do Lung Information Needs Questionnaire (LINQ) depois de uma intervenção educativa em pacientes com doença respiratória crônica]. Revsta Ciencias de la Salud. 15(2): 259-271

10) Crisafulli E, Morandi A, Olivini A, Malerba M, Clini EM. (2014) Rehabilitation and supportive therapy in elderly patients with chronic obstructive pulmonary disease. European Journal of Internal Medicine 25(4): 329-335

11) Spruit MA, Singh SJ, Garvey C, (...), Donner CF, Wouters EFM. (2013) An official American thoracic society/European respiratory society statement: Key concepts and advances in pulmonary rehabilitation. American Journal of Respiratory and Critical Care Medicine

188(8): e13-e64

12) Verbrugge R, , de Boer F, Georges JJ. (2013) Strategies used by respiratory nurses to stimulate self-management in patients with COPD. *Journal of Clinical Nursing*. 22(19-20): 2787-2799

13) Bolton CE, Bevan-Smith EF, Blakey JD, (...), Walker PP, Walmsley S. (2013) British Thoracic Society guideline on pulmonary rehabilitation in adults. *Thorax*. 68(SUPPL. 2): ii1-ii30

14) Cosgrove D, MacMahon J, Bourbeau J, Bradley, J.M., O'Neill, B. (2013) Facilitating education in pulmonary rehabilitation using the Living Well with COPD programme for pulmonary rehabilitation: A process evaluation. *BMC Pulmonary Medicine*. 13(1): 50

15) Paneroni M, Clini, E., Crisafulli E, (...), Bianchi L, Vitacca M. (2013) Feasibility and effectiveness of an educational program in Italian COPD patients undergoing rehabilitation. *Respiratory Care*. 58(2): 327-333

16) Jones RCM, Haskell S. (2012) Know the score: A new tool to help assess, address, and evaluate COPD education. *Canadian Journal of Respiratory Therapy*. 48(1): 26-27

17) Wakabayashi R, Motegi T, Yamada K, (...), Gemma A, Kida K. (2011) Efficient integrated education for older patients with chronic obstructive pulmonary disease using the Lung Information Needs Questionnaire. *Geriatrics and Gerontology International*. 11(4): 422-430

18) Scott AS, Baltzan MA, Dajczman E, Wolkove N. (2011) Patient knowledge in chronic obstructive pulmonary disease: Back to basics. *COPD: Journal of Chronic Obstructive Pulmonary Disease*. 8(5): 375-379

19) Singh S, Harrison S, Houchen L, Wagg K. (2011) Exercise assessment and training in pulmonary rehabilitation for patients with COPD. *European Journal of Physical and Rehabilitation Medicine*. 47(3): 483-497

20) Dodd JW, Hogg L, Nolan J, (...), Man WDC, Hopkinson,NS. (2011) The COPD assessment test (CAT): Response to pulmonary rehabilitation. A multicentre, prospective study. *Thorax* 66(5): 425-429

21) Carlin BW. (2009) Pulmonary rehabilitation and chronic lung disease: Opportunities for the respiratory therapist. *Respiratory Care* 54(8): 1091-1099

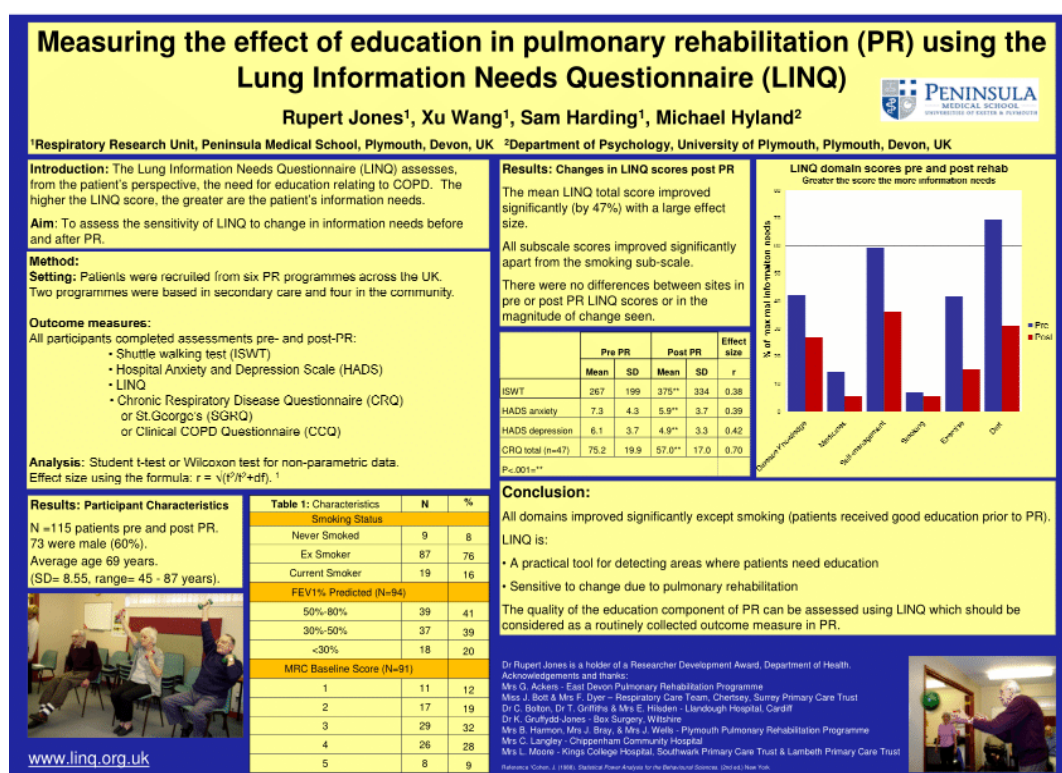
Paper 3, at the time of writing, has a Mendeley statistic of 43.

As with the papers in Chapter 3, Dr Phil Clatworthy and I applied a QA tool. Once again we used the Joanna Briggs Institutes range of tools and for paper 3 we used the Case Series checklist.

Quality Appraisal - Case Series (Moola, Munn, Tufanaru et al, 2017)

	Yes	No	Unclear	Not applicable
1. Were there clear criteria for inclusion in the case series?	☒	☐	☐	☐
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	☒	☐	☐	☐
3. Were valid methods used for identification of the condition for all participants included in the case series?	☒	☐	☐	☐
4. Did the case series have consecutive inclusion of participants?	☐	☒	☐	☐
5. Did the case series have complete inclusion of participants?	☐	☒	☐	☐
6. Was there clear reporting of the demographics of the participants in the study?	☒	☐	☐	☐
7. Was there clear reporting of clinical information of the participants?	☒	☐	☐	☐
8. Were the outcomes or follow up results of cases clearly reported?	☒	☐	☐	☐
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	☒	☐	☐	☐
10. Was statistical analysis appropriate?	☒	☐	☐	☐

Appendix 8: LINQ Poster



Appendix 9: Impact of research of paper 4

Due to the very limited amount of research being undertaken at the time of data collection, I started to disseminate early findings in 2003, with an oral presentation to the British Hyperbaric Association. This was followed up the next year by 2 poster presentations (Appendix 10) and an oral presentation at international conferences and then a further oral presentation (Appendix 11) in 2005 to the Undersea and Hyperbaric Medical Society. Paper 4 was published in the International Journal of Oral and Maxillofacial Surgery. This is the same journal as paper 2. The impact factor, Source Normalized Impact per Paper and abstracting details are presented in section appendix 5, although there is no evidence to indicate that this has changed practice.

In the eleven years since this paper was published it has been cited **33** times.

- 1) Ravi P, Vaishnavi D, Gnanam A, Krishnakumar Raja VB. (2017) The role of hyperbaric oxygen therapy in the prevention and management of radiation-induced complications of the head and neck – a systematic review of literature. Journal of Stomatology, Oral and Maxillofacial Surgery. 118(6): 359-362
- 2) Strojjan P, Hutcheson KA, Eisbruch A, (...), Rinaldo A, Ferlito A. (2017) Treatment of late sequelae after radiotherapy for head and neck cancer. Cancer Treatment Reviews. 59: 79-92
- 3) Olsson AB, Dillon J, Kolokythas A, Schlott BJ. (2017) Reconstructive Surgery. Journal of Oral and Maxillofacial Surgery. 75(8): e264-e301
- 4) Chuinard AF, Giasson L, Fortin M. (2016) Hyperbaric oxygen therapy for head and neck irradiated patients with special attention to oral and maxillofacial treatments. Journal of the Canadian Dental Association. 82: g24
- 5) Curi MM, Cardoso CL, De Lima HG, Kowalski LP, Martins MD. (2016) Histopathologic and histomorphometric analysis of irradiation injury in bone and the surrounding soft tissues of the jaws. Journal of Oral and Maxillofacial Surgery. 74(1): 190-199
- 6) Rogers SN,
D'Souza [https://www.scopus.com/authid/detail.uri?origin=resultslist&auth](https://www.scopus.com/authid/detail.uri?origin=resultslist&authorId=15752582400&zone=)
[orId=15752582400&zone=](https://www.scopus.com/authid/detail.uri?origin=resultslist&authorId=15752582400&zone=) JJ, Lowe D, Kanatas A. (2015) Longitudinal

- evaluation of health-related quality of life after osteoradionecrosis of the mandible. *British Journal of Oral and Maxillofacial Surgery*. 53(9): 854-857
- 7) Van Den Bergh B, De Mol Van Otterloo JJ, Van Der Ploeg T, Tuinzing DB, Forouzanfar T. (2015) IMF-screws or arch bars as conservative treatment for mandibular condyle fractures: Quality of life aspects. *Journal of Cranio-Maxillofacial Surgery*. 43(7): 1004-1009
- 8) Möst T, Schlegel KA, Lutz R, Schmitt C. (2015) At-risk patients in implant dentistry | [Risikopatienten in der implantologie]. *Implantologie*. 23(1): 39-48
- 9) Fox NF, Xiao C, Sood AJ, (...), Sharma A, Day TA. (2015) Hyperbaric oxygen therapy for the treatment of radiation-induced xerostomia: A systematic review. *Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology*. 120(1): 22-28
- 10) Zheng M, Li L, Tang Y, Liang X-H. (2014) How to improve the survival rate of implants after radiotherapy for head and neck cancer? *Journal of Periodontal and Implant Science*. 44(1): 2-7
- 11) Lee A, Forner L, Jansen EC. (2014) Patients perspective on Hyperbaric Oxygen treatment of Osteoradionecrosis. *International Journal of Technology Assessment in Health Care*. 96
- 12) Lee A, Forner L, Jansen EC. (2014) Patients perspective on Hyperbaric Oxygen treatment of Osteoradionecrosis. *International Journal of Technology Assessment in Health Care* 30(2): 188-193
- 13) Erdemci F, Gunaydin Y, Sencimen M, (...), Dogan N, Gider IK. (2014) Histomorphometric evaluation of the effect of systemic and topical ozone on alveolar bone healing following tooth extraction in rats. *International Journal of Oral and Maxillofacial Surgery*. 43(6): 777-783
- 14) Donos N, Calciolari E. (2014) Dental implants in patients affected by systemic diseases. *British Dental Journal*. 217(8): 425-430
- 15) Irgens A, Vaagbo G, Aanderud L. (2013) Quality of life - The effect of hyperbaric oxygen treatment on radiation injury. *Undersea and Hyperbaric Medicine*. 40(6): 479-485
- 16) Singer S, Arraras JI, Chie W-C, (...), Keszte J, Hofmeister D. (2013) Performance of the EORTC questionnaire for the assessment of quality of life in

head and neck cancer patients EORTC QLQ-H&N35: A methodological review. *Quality of Life Research*. 22(8): 1927-1941

17) Jacobson AS, Zevallos J, Smith M, (...), Persky M, Urken ML. (2013) Quality of life after management of advanced osteoradionecrosis of the mandible. *International Journal of Oral and Maxillofacial Surgery*. 42(9): 1121-1128

18) Bassi F, Carr AB, Chang T-L, (...), Stanford CM, Wolfaardt J. (2013) Functional outcomes for clinical evaluation of implant restorations. *International Journal of Prosthodontics* 26(5): 411-418

19) Harding S, Courtney D, Hodder S, Bryson P. (2012) Effects of hyperbaric oxygen therapy on quality of life in Maxillofacial patients with type III osteoradionecrosis. *Journal of Oral and Maxillofacial Surgery*. 70(12): 2786-2792

20) Dunham J. (2012) Reconstructive surgery. *Journal of Oral and Maxillofacial Surgery*. 70(11): e272-e309

21) Danesh-Sani SA, Shariati-Sarabi Z, Feiz MR. (2012) Comprehensive review of hyperbaric oxygen therapy. *Journal of Craniofacial Surgery* 23(5): e483-e491

22) Kuffler DP. (2012) Hyperbaric oxygen therapy: Can it prevent irradiation-induced necrosis? *Experimental Neurology*. 235(2): 517-527

23) Gunepin M, Derache F, Cathelinaud O, (...), Hugon M, Riviere D. (2012) Therapeutic use of hyperbaric oxygen in the prevention of failure of dental implant in irradiated patients | [Intérêt de l'utilisation de l'oxygénothérapie hyperbare en prévention de l'échec implantaire chez le patient irradié]. *Medecine Buccale Chirurgie Buccale*. 18(2): 143-153

24) Vissink A, Spijkervet FKL, Jensen SB, Brennan MT. (2011) Xerostomia. *Supportive Oncology*. pp 232-242.

25) Forner L, Hyldegaard O, Von Brockdorff AS, (...), Nauntofte B, Jensen, S.B. (2011) Does hyperbaric oxygen treatment have the potential to increase salivary flow rate and reduce xerostomia in previously irradiated head and neck cancer patients? A pilot study. *Oral Oncology*. 47(6): 546-551

26) Blochowiak K, Sokalski J. (2011) An application of hyperbaric oxygen therapy in oral and maxillofacial surgery | [Zastosowanie tlenoterapii

hiperbarycznej w chirurgii stomatologicznej i szczekowo-twarzowej]. Dental and Medical Problems. 48(1): 55-60

27) Shaw RJ, Dhanda J. (2011) Hyperbaric oxygen in the management of late radiation injury to the head and neck. Part I: Treatment. British Journal of Oral and Maxillofacial Surgery. 49(1): 2-8

28) Spiegelberg L, Djasim UM, Van Neck HW, Wolvius EB, Van Der Wal KG. (2010) Hyperbaric oxygen therapy in the management of radiation-induced injury in the head and neck region: A review of the literature. Journal of Oral and Maxillofacial Surgery. 68(8): 1732-1739

29) Jensen SB, Pedersen AML, Vissink A, (...), Spijkerver FKL, Brennan MT. (2010) A systematic review of salivary gland hypofunction and xerostomia induced by cancer therapies: Management strategies and economic impact. Supportive Care in Cancer. 18(8): 1061-1079

30) Thariat J, de Mones E, Darcourt V, (...), Bensadoun RJ, Dassonville O. (2010) Teeth and irradiation: Dental care and treatment of osteoradionecrosis after irradiation in head and neck cancer | [Dent et irradiation: prévention et traitement des complications dentaires de la radiothérapie y compris l'ostéoradionécrose]. Cancer/Radiothérapie. 14(2): 137-144.

31) Kanatas AN, Rogers SN. (2010) A systematic review of patient self-completed questionnaires suitable for oral and maxillofacial surgery. British Journal of Oral and Maxillofacial Surgery. 48(8): 579-590

32) Goiato MC, Santos DM, Danelon M, (...), De Carvalho Dekon SF, Fajardo RS. (2009) Hyperbaric oxygen: Therapy for patients with maxillofacial implants? Journal of Craniofacial Surgery. 20(5): 1519-1522

33) Vissink A, Raghoobar GM, Roodenburg JLN, (...), Burge FR, Langendijk JA. (2009) Does hyperbaric oxygen therapy improve quality of life? International Journal of Oral and Maxillofacial Surgery. 38(1): 99-100

Quality Appraisal - Case Series (Moola, Munn, Tufanaru et al, 2017)

	Yes	No	Unclear	Not applicable
1. Were there clear criteria for inclusion in the case series?	☒	☐	☐	☐
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	☒	☐	☐	☐
3. Were valid methods used for identification of the condition for all participants included in the case series?	☒	☐	☐	☐
4. Did the case series have consecutive inclusion of participants?	☒	☐	☐	☐
5. Did the case series have complete inclusion of participants?	☒	☐	☐	☐
6. Was there clear reporting of the demographics of the participants in the study?	☒	☐	☐	☐
7. Was there clear reporting of clinical information of the participants?	☐	☒	☐	☐
8. Were the outcomes or follow up results of cases clearly reported?	☒	☐	☐	☐
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	☐	☒	☐	☐
10. Was statistical analysis appropriate?	☒	☐	☐	☐

Appendix 10: Hyperbaric Oxygen Therapy and Quality of Life Poster



An investigation into the impact of hyperbaric oxygen therapy on quality of life in patients with maxillofacial soft tissue- & osteo- radionecrosis

Harding SA, Courtney DJ*, Hodder SJ†, Bryson PJ.
 DDRc, Hyperbaric Medical Centre, Derriford, Plymouth, Devon, PL6 8BU, UK
 *Dept of Oral & Maxillofacial Surgery, Derriford Hospital, Derriford, Plymouth, Devon, PL6 8BU, UK
 † Dept of Oral & Maxillofacial Surgery, Morriston Hospital, Morriston, Swansea, SA6 6NL, UK

Introduction:

A lack of research into Quality of Life (QoL) changes in patients with maxillofacial soft tissue- and osteo-radionecrosis who undergo hyperbaric oxygen therapy has led to a knowledge gap. This study aims to fill the knowledge base for the use of HBO₂ within the maxillofacial community.

Methodology:

2004, patients referred consecutively for HBO₂ following radiotherapy to head and neck cancers were asked to complete questionnaires pre & post HBO₂ (part 1 of a 2 part study).

Ethical approval was granted from the Local Research Ethics Committee and patient consent obtained.

Four standardised validated questionnaires were used:

- Global non disease specific
- Hospital Anxiety and Depression Scale (HADS) (see score + low levels of anxiety and depression)
- Medical Outcomes Study Short Form-36 (SF-36) (see score + poor health)

Disease specific scales:

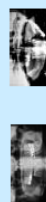
- University of Washington - QOL (UW-QOL) (see score + poor health)
- University of Washington - Pain (UW-Pain) (see score + poor health)
- University of Washington - Swallowing (UW-Swallowing) (see score + poor health)
- University of Washington - Dry Mouth (UW-Dry Mouth) (see score + poor health)

The supplementary module QLQ-HAMS (HAMS) (see score + poor health) including the supplementary module QLQ-HAMS (HAMS) (see score + poor health) post-HBO₂.

The questionnaire battery was given at two time points: pre and immediately post-HBO₂.

Therapies were administered at 2.2 ATA (two periods of 45 minutes) twice a day.

Scores between each time point were compared using Paired Samples T-Tests.

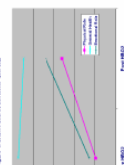


Results:

59 patients were recruited and completed the pre and post therapy questionnaires. The study group consisted of 46 males and 13 females (mean age 57 years). Patients received 23 to 38 HBO₂ therapies each (mean = 30, SD = 4.3).

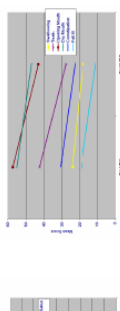
No significant changes were identified in either category (Anxiety or Depression) using the HADS. The SF-36 showed significant changes in three of the eight categories: general health declined (P=0.031), physical role (P=0.001) and emotional functioning (P=0.001) improved, and these trends are shown in Fig. 1.

Figure 1: SF-36 Physical Role (mean score over time)



Only one of the twelve categories in the UW-QOL scale was significant, showing a positive improvement in taste (P=0.029). Two out of the three categories in the UW-Pain scale were significant, showing a positive improvement in pain (P=0.001) and pain (P=0.001). The UW-Swallowing scale showed a significant positive change in swallowing (P=0.003). The UW-Dry Mouth scale showed a significant positive change in dry mouth (P=0.003). The QLQ-HAMS scale showed a significant positive change in swallowing (P=0.003), teeth (P=0.003), opening mouth (P=0.001), dry mouth (P=0.003), constipation (P=0.003), and are illustrated in Fig. 2.

Figure 2: QLQ-HAMS (mean score over time)



Discussion:

Although this study population was drawn from routine referrals and conducted over standard therapies, and therefore included patients with differing primary diagnoses, the results show a positive impact on QoL following HBO₂ therapies.

Treatment for Head and Neck cancer results in short term morbidity, most of which resolves within 1 year. Despite an initially high level of depressive symptomatology, there is gradual improvement of psychological functioning and global QoL over the course of three years. Previous research with this type of patient has shown that there is a resilience to gradual improvement, for example senses (taste/smell), dry mouth, sticky saliva & sexuality. Many of the patients in our study are more than three years post cancer treatment and should therefore have stabilised QoL. The gradual improvement may be due to the HBO₂ therapies or a combination of HBO₂ and Surgery.

Some of the changes in this study, Physical Role and Global Health Status, are functional in nature. Other improvements reported are in the Symptoms Categories: Pain, Swallowing, Opening Mouth, Dry Mouth, Teeth, Constipation and less Feelings of Illness. However, analysis in relation to general functioning (SF-36) showed a decline in physical role and emotional functioning. The EORTC QLQ-C30 measures cancer related QoL. The lack of negative changes in QoL during the patients treatment is reassuring.

Of note is that the more long term detrimental symptoms, widely regarded as difficult to treat and experienced by this type of patient (e.g. Trismus and Xerostomia) improved. This identifies a possible benefit from HBO₂.

The study was not designed to test QoL in its broadest sense (e.g. happiness, personal goals). A broader based assessment would be profitable at a time post-treatment requiring complex methodology.

Conclusions:

Patients in this study group showed some important changes in QoL after receiving HBO₂ therapies identified within the questionnaire battery.

This study is part 1 of a longitudinal project designed to assess the changes in QoL over a year post HBO₂.

2022-01-01 10:00:00 - 2022-01-01 10:00:00

Appendix 11: Hyperbaric Oxygen Therapy and Quality of Life Presentation

An Improved Quality of Life in Patients Receiving Hyperbaric Oxygen Therapy as a Conservative Treatment with Maxillofacial Radiation Tissue Damage

The Ameliorating Effects of Hyperbaric Oxygen Therapy on Quality of Life in Patients with Maxillofacial Radiation Tissue Damage

Harding SA, Courtney DJ*, Hodder SC†, Bryson PJ.

DDRC, Hyperbaric Medical Centre, Plymouth, Devon, UK.

*Dept of Oral & Maxillofacial Surgery, Derriford Hospital, Plymouth, Devon, UK.

†Dept of Oral & Maxillofacial Surgery, Morriston Hospital, Morriston, Swansea UK



- 6830 people in 2002 were registered in the UK as having head and neck cancers
 - Approximately 1.5% develop long term complications related to radiotherapy
 - HBO₂ has been identified as a useful adjunctive therapy for radiation tissue damage (Feldmeier, J.J. 2004)
 - Studies using this group of patients having HBO₂, have not fully considered changing Quality of Life (QoL)
- QoL is becoming increasingly relevant to professional judgement with regard to therapeutic regimes
- Currently there are no hyperbaric specific QoL measures
- How do you measure QoL?



- **Global Scales** - "individual's perception of their position in life in the context of the culture and value system in which they live and in relation to their goals, expectations, standards and concerns" (The World Health Organization)
- **Cancer Specific Scales** - individual's perception of their position in life in the context of their experience and functionality due to cancer
- **Head & Neck Cancer Scales** - individual's perception of their position in life in relation to their function and symptoms having (had) head and neck cancer



Methodology

Global Questionnaires	General Cancer	Head & Neck Cancer
HADS	EORTC C30	EORTC H&N35
SF-36		UoW

Time Points

Conservative Group	Peri Operative Group
Assessed	Assessed
HBO ₂ (Mean 35)	HBO ₂ (Mean 26)
Assessed	Surgery
	HBO ₂ (Mean 17)
	Assessed



Results:

Conservative Group		Sample Size
		27
Peri Operative Group	Debridement	24
	Extraction or Implant	49
All Participants – Total		100

Using the Global scale 'HADS' - No significant differences in Anxiety or Depression were identified in any group



Global QoL Scales

Categories showing significant changes

SF- 36

	Conservative	Debridement	Extraction or Implants	All Participants
Role-Physical	x	x	x	P≤0.020
Bodily Pain	x	x	P≤0.034	x
Social Functioning	P≤0.033	x	x	x
Role-Emotional	P≤0.033	x	x	P≤0.007

Cancer Specific Questionnaires

EORTC C30

	Conservative	Debridement	Extraction or Implants	All Participants
Emotional	x	x	x	P≤0.015
Pain	x	x	P≤0.005	P≤0.021
Global Health	x	x	P≤0.016 ↓	P≤0.015
Dyspnoea	x	P≤0.009	x	P≤0.013

x = No Significant Differences, ↓ = Decline in QoL



Head and Neck Specific Questionnaire

EORTC H&N35	Conservative	Debridement	Extraction or Implants	All Participants
Swallowing	P≤0.001	x	x	P≤0.006
Senses Problems	P≤0.050	x	x	x
Trouble with Social Eating	P≤0.007	x	x	x
Less Sexuality	x	x	x	P≤0.043 ↓
Teeth	P≤0.026	x	P≤0.001	P≤0.001
Opening Mouth	x	x	P≤0.047	P≤0.020
Dry Mouth	x	x	x	P≤0.008

UoW	Conservative	Debridement	Extraction or Implants	All Participants
Pain	x	x	P≤0.034	x
Chewing	x	x	P≤0.044 ↓	x
Taste	x	P≤0.040	x	P≤0.034
Saliva	P≤0.023	x	x	P≤0.006

x = No Significant Differences, ↓ = Decline in QoL



Discussion:

- Analysis of the data shows significant changes
- Questionnaires were selected from valid, reliable and sensitive measures
- Population drawn from routine referrals
- A range of neoplasm sites
- Standard HBO₂ therapies
- Self-selecting patients



Discussion Continued

- Information can be used by referring consultants
- Consultants can consider HBO₂ as a treatment for the long term effects of radiotherapy (e.g. Xerostomia & Trismus)
- HBO₂ has additional advantages not taken into account
- Assess functional outcomes



Discussion Continued

- Information can be used by referring consultants
- Consultants can consider HBO₂ as a treatment for the long term effects of radiotherapy (e.g. Xerostomia & Trismus)
- HBO₂ has additional advantages not taken into account
- Assess functional outcomes

Thank you
Questions?



Appendix 12: Impact of research of paper 5

As with paper 4, there was a very limited amount of research being undertaken at the time of data collection. I started to disseminate early findings in 2004, through an oral presentation to the British Hyperbaric Association. This was followed the next year by two poster presentations and an oral presentation at international conferences and then a further oral presentation in 2005 to the Undersea and Hyperbaric Medical Society. Paper 5 was published in the Journal of Oral and Maxillofacial Surgery. This is the American Association of Oral and Maxillofacial surgeons' journal and at the time of publishing this article the impact factor was 1.78 and Source Normalized Impact per Paper value was 1.45. In comparison to paper 4 this has been cited fewer times, **7** as compared to 34 (at the time of writing). This is likely due to the rarity of the disease in combination with the use of Hyperbaric Oxygen Therapy. Meaning that the reference is less likely to be cited by other researchers, as the patient cohort is hard to access.

- 1) Strojan P, Hutcheson KA, Eisbruch A, (...), Rinaldo A, Ferlito A. (2017) Treatment of late sequelae after radiotherapy for head and neck cancer. Cancer Treatment Reviews. 59: 79-92
- 2) Long Y, Tan J, Nie Y, (...), Mei X, Tu C. (2017) Hyperbaric oxygen therapy is safe and effective for the treatment of sleep disorders in children with cerebral palsy. Neurological Research. 39(3): 239-247
- 3) Gottrup F, Dissemond J, Baines C, (...), Krønger K, Longobardi P. (2017) Use of oxygen therapies in wound healing: Focus on topical and hyperbaric oxygen treatment. Journal of Wound Care. 26(5): S1-S44
- 4) Gottrup F, Dissemond J, Baines C, (...), Krønger K, Longobardi P. (2017) Use of oxygen therapies. Journal of Wound Care. 26, pp. S1-S42
- 5) Rogers SN, D'Souza JJ, Lowe D, Kanatas A. (2015) Longitudinal evaluation of health-related quality of life after osteoradionecrosis of the mandible. British Journal of Oral and Maxillofacial Surgery. 53(9): 854-857
- 6) Fox NF, Xiao C, Sood AJ, (...), Sharma A, Day TA. (2015) Hyperbaric oxygen therapy for the treatment of radiation-induced xerostomia: A systematic review. Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology. 120(1): 22-28

- 7) Lee A, Forner L, Jansen EC. (2014) Patients perspective on Hyperbaric Oxygen treatment of Osteoradionecrosis. International Journal of Technology Assessment in Health Care. 96
- 8) Lee A, Forner L, Jansen EC. (2014) Patients perspective on Hyperbaric Oxygen treatment of Osteoradionecrosis. International Journal of Technology Assessment in Health Care. 30(2): 188-193
- 9) Perdrizet GA. (2014) Principles and practice of hyperbaric medicine: A medical practitioner's primer, Part II. Connecticut Medicine. 78(7), pp. 389-402
- 10) Irgens A, Vaagbo G, Aanderud L. (2013) Quality of life - The effect of hyperbaric oxygen treatment on radiation injury. Undersea and Hyperbaric Medicine. 40(6): 479-485
- 11) Forner L (2013) Does hyperbaric oxygen have the potential to improve quality of life in irradiated patients? What is the evidence? An editorial perspective. Undersea and Hyperbaric Medicine. 40(6): 475-477

Quality Appraisal - Case Series (Moola, Munn, Tufanaru et al, 2017)

	Yes	No	Unclear	Not applicable
1. Were there clear criteria for inclusion in the case series?	☒	☐	☐	☐
2. Was the condition measured in a standard, reliable way for all participants included in the case series?	☒	☐	☐	☐
3. Were valid methods used for identification of the condition for all participants included in the case series?	☒	☐	☐	☐
4. Did the case series have consecutive inclusion of participants?	☐	☐	☒	☐
5. Did the case series have complete inclusion of participants?	☐	☐	☒	☐
6. Was there clear reporting of the demographics of the participants in the study?	☒	☐	☐	☐
7. Was there clear reporting of clinical information of the participants?	☒	☐	☐	☐
8. Were the outcomes or follow up results of cases clearly reported?	☒	☐	☐	☐
9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information?	☒	☐	☐	☐
10. Was statistical analysis appropriate?	☒	☐	☐	☐

Appendix 13: Impact of research of paper 6

This paper was published early online towards the end of 2017 and in hard copy in 2018. This paper was a direct development of paper 2 (chapter 3).

The International Journal of Oral and Maxillofacial Surgery at the time of writing has an impact factor of 2.164, and a Source Normalized Impact per Paper value of 1.254. It is currently ranked 24/200 in journals of dentistry, oral surgery and medicine and 77/200 of surgical journals. International Journal of Oral and Maxillofacial Surgery is the journal of the International Association of Oral and Maxillofacial Surgeon. As such International Journal of Oral and Maxillofacial Surgery is available free to International Association of Oral and Maxillofacial Surgeon members. Importantly International Journal of Oral and Maxillofacial Surgery is indexed on some of the key databases (Current Contents / Clinical Medicine, EMBASE/Excerpta Medica, Medline/Index Medicus, Medical Documentation Service, Research Alert, ISI Science Citation Index, SciSearch, BIOSIS/Biological Abstracts, Scopus), ensuring it can be found by a comprehensive range of professionals including medics, scientists, nurses, allied health professionals and researchers.

As research into PPC within a HNC population is a slow moving field, with only 6 other papers being published since 2008 and prior to this one, it is unsurprising it has yet to be cited. However, as with the previously discussed research, I have undertaken other dissemination activities.

An early iteration of the work was presented at the 10th International Conference Head Neck Cancer Quality of Life in 2014. The audience for this conference was a mixture of medics, nurses and allied health professionals, most speech and language therapists. In 2018, I was invited to present the research as published (Appendix 15) with a greater focus on the underlying theory at the Royal College of Speech and Language Therapists Head and Neck Cancer Clinical Excellence Network.

As with paper 2, and mentioned above, this paper has only been in circulation for 18 months and is investigating specialised area of practice. So a Mendeley statistic of 12 is low, but comparable to that of paper 2.

As with all previous paper in this thesis Dr Phil Clatworthy and I applied a QA tool. This time the tool was selected for its suitability for the methodology used within the research.

Quality Appraisal - Cross-Sectional (Moola, Munn, Tufanaru et al 2017)

	Yes	No	Unclear	Not applicable
1. Were the criteria for inclusion in the sample clearly defined?	☒	☐	☐	☐
2. Were the study subjects and the setting described in detail?	☒	☐	☐	☐
3. Was the exposure measured in a valid and reliable way?	☒	☐	☐	☐
4. Were objective, standard criteria used for measurement of the condition?	☒	☐	☐	☐
5. Were confounding factors identified?	☒	☐	☐	☐
6. Were strategies to deal with confounding factors stated?	☒	☐	☐	☐
7. Were the outcomes measured in a valid and reliable way?	☒	☐	☐	☐
8. Was appropriate statistical analysis used?	☒	☐	☐	☐

Appendix 14: Paper 6 Oral presentation



Bristol Speech and
Language Therapy
Research Unit

NHS
North Bristol
NHS Trust

Positive psychological change following head and neck cancer

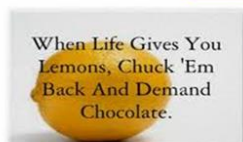
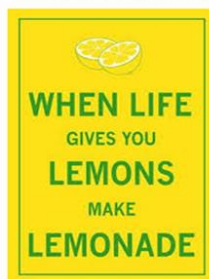
Dr Sam Harding DHealthPsych
Senior Research Associate

Monday 4th June 2018



Public understanding / Proverb driven

“What doesn’t kill you makes you stronger”

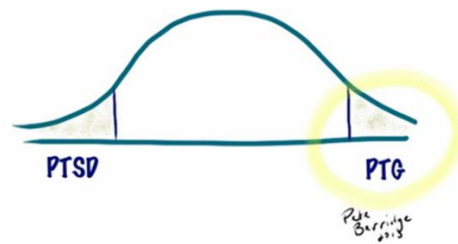


Positive Psychological Change

PPC Synonyms

- Post-traumatic growth
- Perceived benefits
- Benefit finding
- Thriving
- Stress-related growth
- Adversarial growth
- Existential growth

Post Traumatic Stress Disorder & Post Traumatic Growth



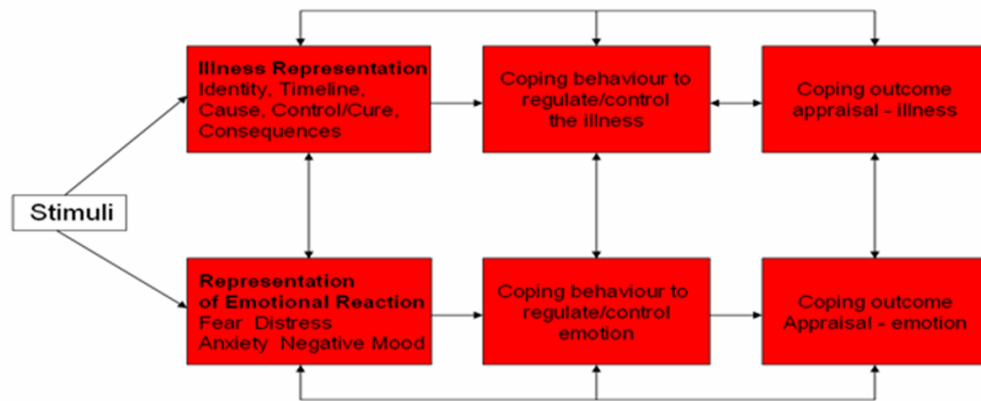
Adversity hypothesis states

“that people can use adversity, trauma and setbacks in order to grow, find fulfilment, develop as a person and find their inner strength”

- Among cancer survivors, there is evidence indicating that a substantial number of people experience such positive changes, especially in the long term.



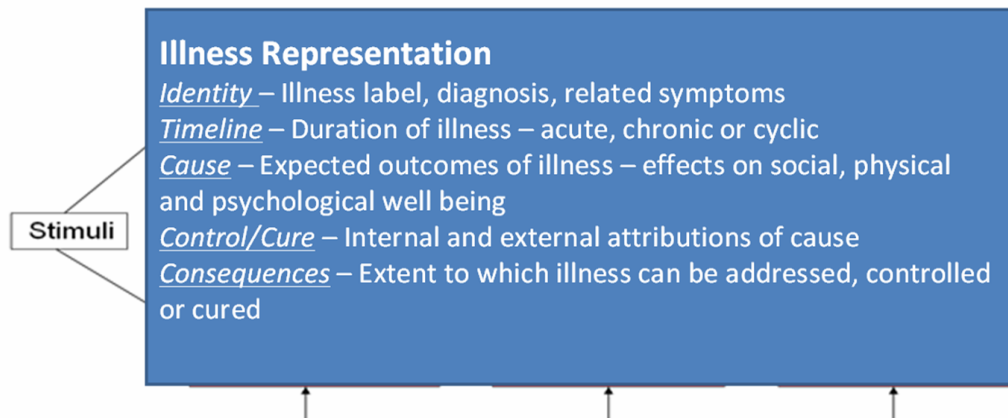
Self-regulatory model of illness behaviour



Leventhal et al., 1984



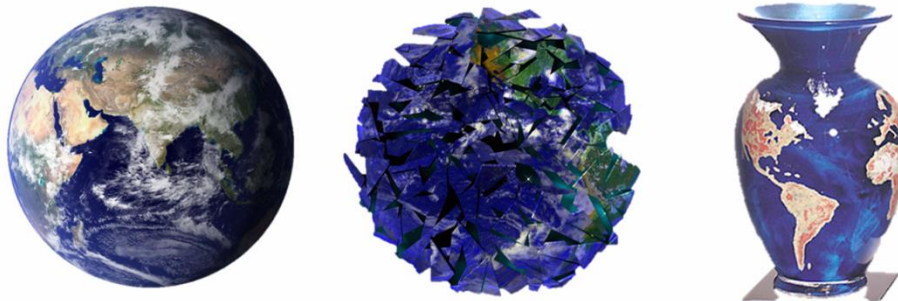
Self-regulatory model of illness behaviour



Leventhal et al., 1984



Assumptive World

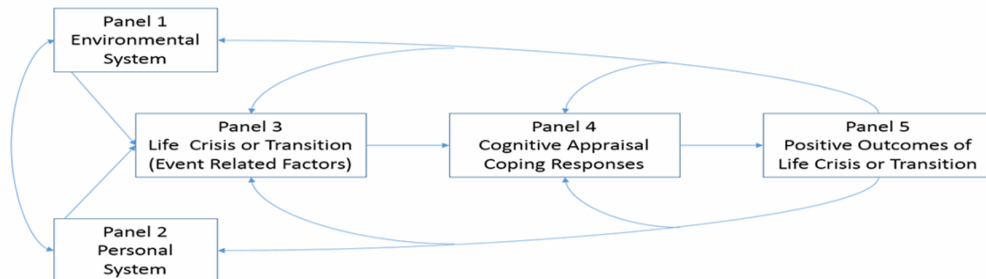


Discussion – Challenges

- When do you collect baseline data?
 - What are pre-trauma variables, including pre-trauma psychological symptoms and resources, that predict PPC?
 - What study design do you use?
- How can you tell if the rehabilitation provided is the source of any PPC development?
 - Natural recovery
 - Pre-existing psychological resilience



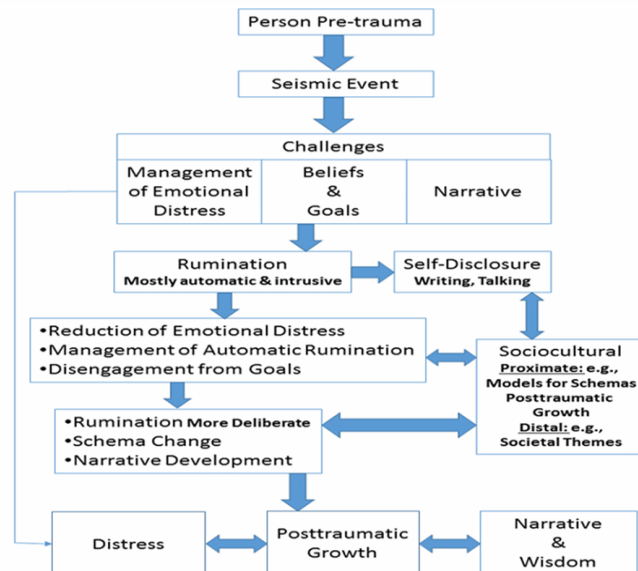
Models of PPC (1)



Life Crisis and Personal Growth (Schaefer and Moos, 1992)



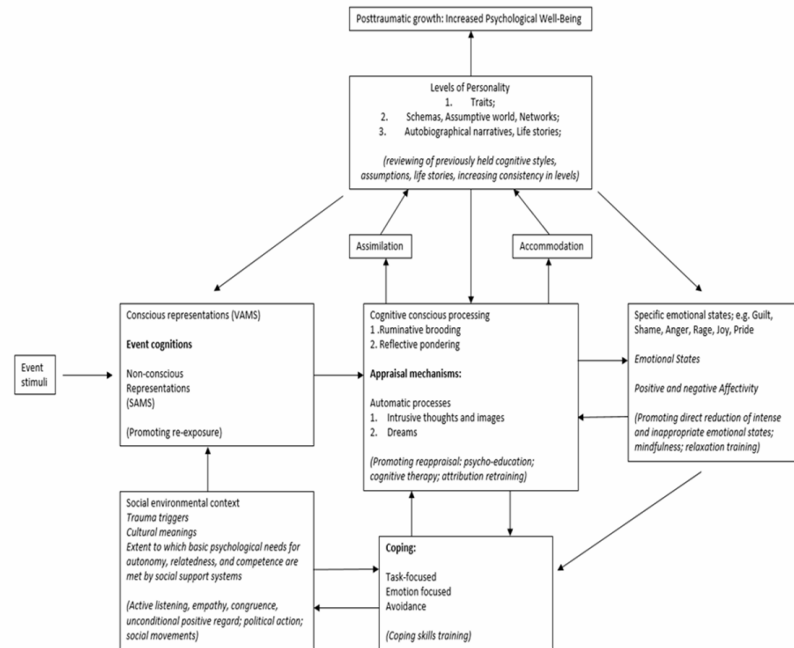
Models of PPC (2)



Comprehensive model of PTG (Tedeschi and Calhoun, 2004)



Models of PPC (3)



Affective-Cognitive Processing Model for Post-Traumatic Growth (Joseph et al. 2012)

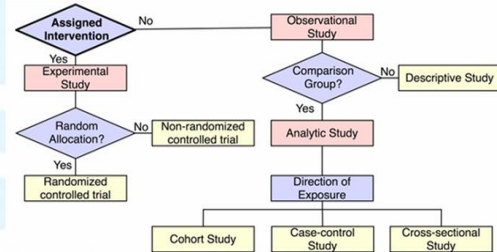
Measures of PPC

- Adult hope scale
- Attributional style questionnaire
- **Benefit finding scale**
- Changes in outlook questionnaire
- Curiosity and exploration inventory
- Gratitude questionnaire
- Inspiration scale
- Meaning in life questionnaire
- Perceived benefits scale
- Personal growth initiative scale
- **Post traumatic growth inventory**
- Psychological well-being scale
- Satisfaction with life scale
- **Silver lining questionnaire**
- State-trait cheerfulness inventory
- Stress-related growth scale
- Subjective happiness scale
- Thriving scale

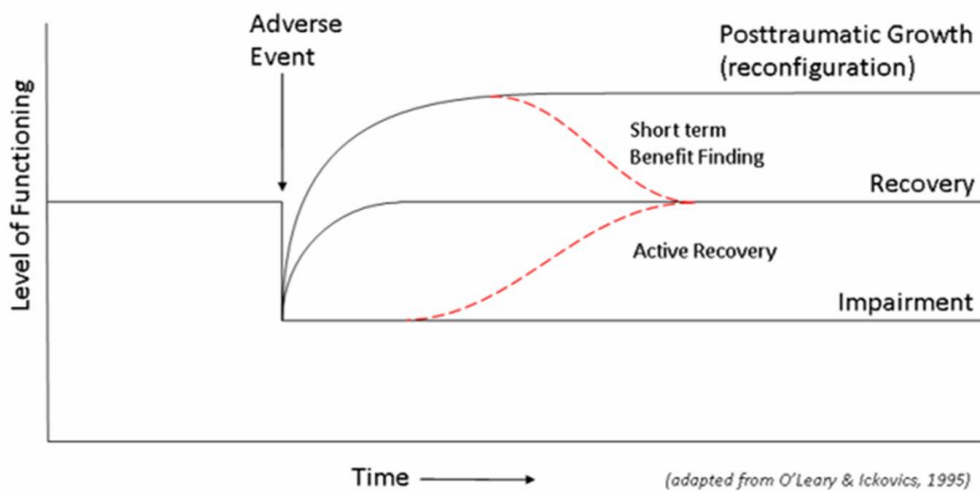
Study Designed used in HNC studies

- Data Collection time points

- Cross-sectional (6 studies)
 - > 1mth post treatment
 - > 3mths post treatment
 - > 6mths post treatment
 - > 6mths post diagnosis
 - > 8mths post treatment
 - < 3 years post diagnosis and post surgery
- Repeated measures (2 studies)
 - Between diagnosis and treatment AND 6 months post treatment
 - Within 1 year of diagnosis AND 6 months following first measure
- Cross sequential (1 study)
 - > 3mths post treatment



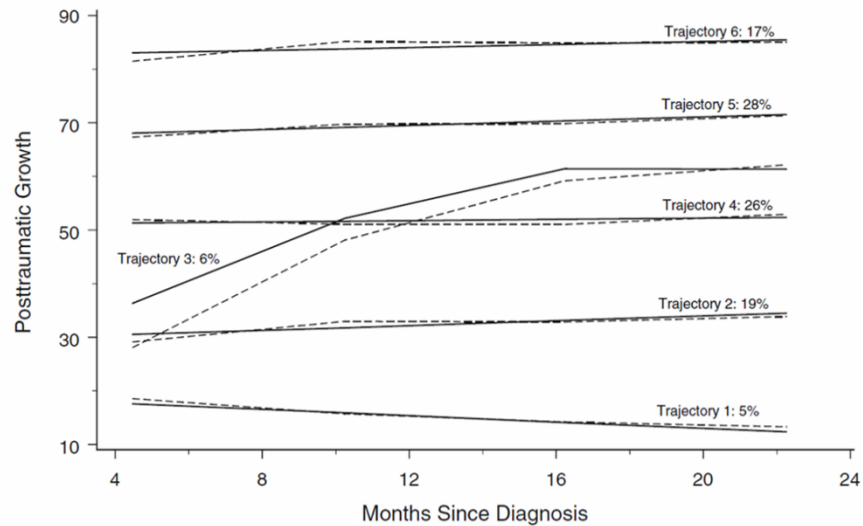
Patterns of PPC development





Trajectories in Breast Cancer Cohort

PTGI total score trajectories in women with Breast Cancer (Danhauer et al., 2015)

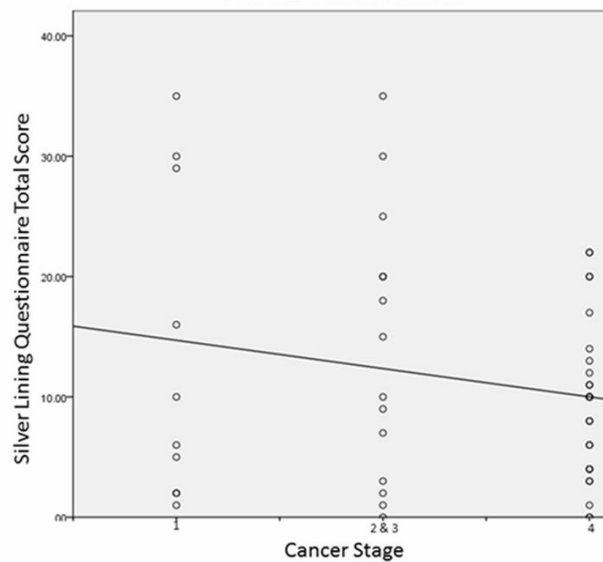


Key: Y-axis shows the PTGI total score trajectories. Percentages shown are the probabilities of group membership. Possible PTGI scores range from 0 to 105. *Dashed lines* reflect observed values, and *solid lines* reflect predicted values.



HNC Stage and PPC (3-12mths)

Silver Lining Questionnaire against Cancer Stage
between 3 and 12 months

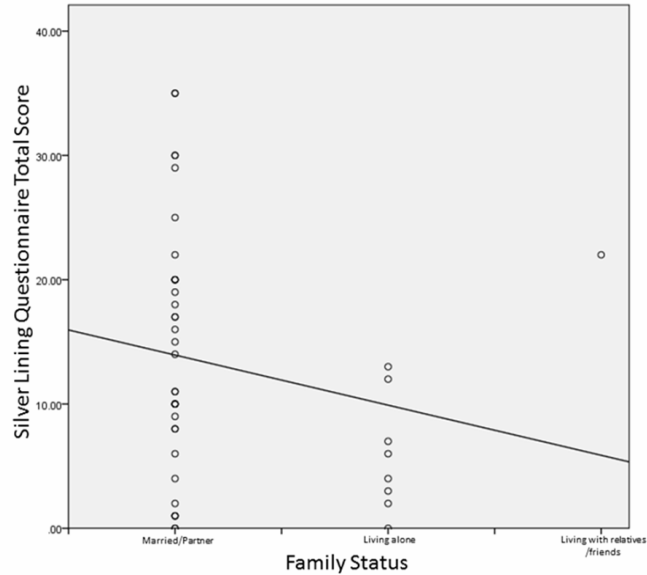


Harding, 2017



Family Status and PPC (3-12mths)

Silver Lining Questionnaire against Family Status between 3 and 12 months

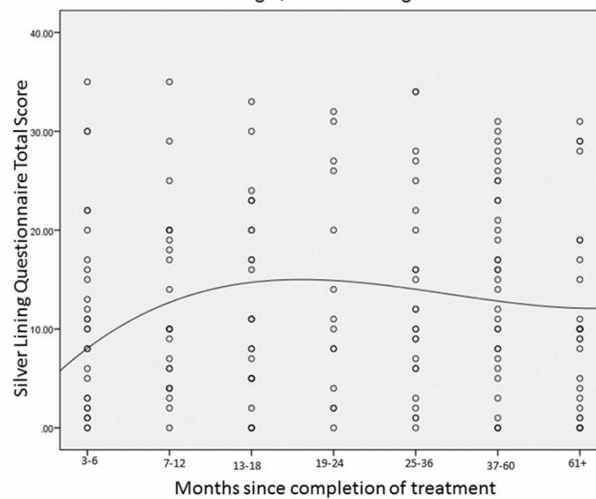


Harding, 2017



Time since treatment and PPC

Silver Lining Questionnaire against Time



Harding, 2017



Discussion

- What is the traumatic experience of HNC?
 - Is it an acute or chronic experience?

	Acute disease	Chronic illness
Onset	Abrupt	Usually gradual
Duration	Limited	Length, indefinite
Cause	Usually single	Usually multiple and changes over time
Diagnosis and Prognosis	Usually accurate	Often uncertain
Technological Intervention	Usually effective	Often indecisive, adverse effects common
Outcome	Cure	No cure
Uncertainty	Minimal	Pervasive
Knowledge	Professionals knowledgeable; patients inexperienced	Professional and patients have complementary knowledge

From Holman and Lorig (2000), Data Supplement – Box, electronic, British Medical Journal (reprinted by kind permission of authors and British Medical Journal, copyright agreement appendix 14)

Discussion

- Current definitions of PPC are insufficiently robust and this means that no single measure directly measures the domains of PPC as described
 - Perceived changes in self
 - Changes in interpersonal relationship
 - Changes in spirituality or finding deeper meaning in life
- Additional research is needed to;
 - Identify if there are multiple longitudinal patterns of development
 - If there is a difference between an acute and chronic events triggering PPC
 - Identification of personality, social, and medical factors that may influence or predict correlations

Appendix 15: Mediators and Moderators of PPC

The research undertaken for paper 6 has been interested in answering the question of how PPC develops or is maintained over an extended time frame. Longitudinal research is not the only area that has been identified as requiring attention in this field of study. The identification and statistical analysis of mediating and moderating factors still needs addressing (Joseph and Linley, 2008).

No research has been published that looks at mediator and moderators of PPC within an HNC population, although work has been undertaken in other areas. Pre-trauma mental health has been identified by Calhoun and Tedeschi (2006) as important for PPC. They predicted that, in line with some other theorists (e.g. Miller and C'deBaca, 1994) "people who experience Posttraumatic Growth need to have had room to grow, but be healthy enough to cope relatively successfully with their emotional distress" (Calhoun and Tedeschi, 1998 p.226). This is the point in which coping plays a part in their model, as those who have adequate coping skills will it predicts, avoid being overwhelmed by trauma, and are more likely to experience PPC.

In a meta-analysis Helgeson *et al.*, (2006) assesses the time that had passed since the trauma. They observed that PPC after a trauma was more strongly related to less depression and more positive affect when the trauma occurred more than two years previously. However, they found that overall time since trauma was not related to an increase in PPC. It could be suggested that time may still be a contributing factor and further research could examine this, as out of the 77 articles that Helgeson *et al.* (2006) reviewed, only 6 (8%) reported data concerning the time that had passed since the incident occurred. It is also the case that the variation in the time points makes it difficult to make an accurate comment on how time affects the development of PPC. The curvilinear effect of time since diagnosis on PPC development in the current study suggests that time may also be a complicating factor.

The relationships of cancer stage and treatment regimen with PPC found in my work would be an interesting investigation. As it may be that treatment regimens have a moderating effect on the mediator, cancer stage.

Patient and Clinicians Perspectives of HNC

I would also be interested to undertake future research that may elicit understanding of the patient's perspective. This research would examine patient and clinician interactions, and then subsequently what each actor thought independently about the trauma of cancer and the development of PPC. This would allow for insights into the description of the disease, treatment, outcomes as well as the short and long term expectations and beliefs about the HNC disease journey. This is important as the understanding of an illness may affect the psychological impact of a treatment and its side effects.

A key consideration in, my opinion, which needs illumination is whether HNC is considered to be acute or chronic. It has been suggested that patients with a chronic condition may be expected to adopt a more active role in managing their problems than might be expected from those with an acute medical problem (White, 2001). Holman and Lorig (2000) have highlighted some differences between acute and chronic illnesses and comparison of the definitions offers a challenge to understanding cancer using these headings. It is likely that some people with cancer had no idea that they are going to receive a cancer diagnosis and therefore it can be argued that the disease onset is abrupt, but others may have suspected that this diagnosis was likely and in some cases have delayed seeking medical intervention and lived with the gradually increasing symptoms. Additionally, how the prognosis is described to the patient may well have a lasting impact on when the patient thinks they have 'come out of the other side' of their illness. One patient may believe that on completion of their surgery they have overcome the disease, whilst another may not feel like this until they are discharged from the service five years after they have completed their treatment. Indefinite outcomes and the associated pervasive levels of uncertainty surrounding diagnosis and prognosis, as well as the side effects of treatment may be central to patients' perceptions and development of PPC. Accepting the ambiguity of the triggering event may be crucial for cancer survivors.

Positive Psychological Change Intervention(s)

Park and Helgeson (2006) have cautioned against the rapid development of large-scale PPC interventions among individuals who have experienced a traumatic life event before a number of key conceptual and empirical questions are answered, in particular, the time course of PPC development. That is, at what point does a coping strategy used by an individual in an acutely stressful period solidify into a cognitive and behavioural change?

If initial reports of positive change are short term coping or, as discussed above, a form of responding in a socially desirable way, this could compromise the rehabilitation process as, for example, cancer survivors reporting such PPC may be less likely to engage with rehabilitation services or attend hospital review appointments.

Paper 6 shows longitudinal research which starts to shine a light on the pattern of PPC development in people who have been treated for HNC. However, a greater understanding of the development and trajectories of change, including variables that differentiate sub-groups, is required before it would be possible to comment on whether an intervention had successfully impacted on the development of PPC.

Appendix 16 Measure of Positive Psychological Change

Questionnaire Title	Questionnaire Title Abbreviation	Self-Report	Number of items	Response Format	Completion Time	Other Details	Key References
Adult Hope Scale	AHS	Yes	12	8-point Likert-type scale	Few Minutes	The adult hope scale (AHS) measures Snyder's cognitive model of hope which defines hope as "a positive motivational state that is based on an interactively derived sense of successful (a) agency (goal-directed energy), and (b) pathways (planning to meet goals)".	1. Snyder, C. R., Harris, C., Anderson, J. R., Holleran, S. A., Irving, L. M., Sigmon, S. T., et al.(1991). The will and the ways: Development and validation of an individual-differences measure of hope. <i>Journal of Personality and Social Psychology</i> , 60, 570-585. 2. Snyder, C. R. (1994). <i>The psychology of hope: You can get there from here</i> . New York: Free Press. 3. Snyder, C. R. (2002). Hope theory: Rainbows in the mind. <i>Psychological Inquiry</i> , 13, 249-275.
Attributional Style Questionnaire	ASQ	Yes	12	7-point Likert-type scale	20 Minutes	Yields scores for explanatory style for bad events and for good events using three causal dimensions: internal versus external, stable versus unstable, and global versus specific causes.	1. Buchanan, G. and Seligman, M.E.P. (Eds.). (1995). <i>Explanatory Style</i> . Hillsdale, N.J.: Erlbaum. 2. Peterson, C. (1988). Explanatory style as a risk factor for illness. <i>Cognitive Therapy and Research</i> , 12, 117-130. 3. Peterson, C., Semmel, A., von Baeyer, C., Abramson, L. T., Metalsky, G. I., & Seligman, M. E. P. (1982). The Attributional Style Questionnaire. <i>Cognitive Therapy and Research</i> , 6, 287-300. 4. Seligman, M.E.P., Nolen-Hoeksema, S., Thornton, N., and Thornton, K.M. (1990). Explanatory style as a mechanism of disappointing athletic performance. <i>Psychological Science</i> , 1, 143-146. 5. Seligman, M.E.P. and Schulman, P. (1986). Explanatory style as a predictor of productivity and quitting among life insurance agents. <i>Journal of Personality and Social Psychology</i> , 50, 832-838. 6. Sweeney, P.D., Anderson, K, & Bailey, S. (1986). Attributional style in depression: A meta-analytic review. <i>Journal of Personality and Social Psychology</i> , 50, 974-991.

Questionnaire Title	Questionnaire Title Abbreviation	Self-Report	Number of items	Response Format	Completion Time	Other Details	Key References
Benefit Finding Scale	BFS	Yes	17	5 point Likert type scale	Not stated	Items were adapted from Behr's Positive Contributions scale used with parents of disabled children (Behr, Murphy & Summer, 1991). Psychometrics are established with 364 breast cancer patients. Cronbachs alpha is 0.95	1. Tomich, P. L., & Helgeson, V. S. (2004). Is finding something good in the bad always good? Benefit finding among women with breast cancer. <i>Health Psychology</i> , 23, 16-23. 2. Behr, S.K., Murphy, D.L., & Summer, J.A. (1991). <i>Kansas inventory of parental perceptions</i> . Lawrence: University of Kansas. 3. Carver, C.S. (2013) The Benefit Finding Scale for breast cancer. Measurement Instrument Database for the Social Science. Retrieved from www.midss.ie .
Changes in Outlook Questionnaire	CiOQ	Yes	26	6 point Likert type scale	Not stated	Internal consistency reliability of the positive scale is -0.79 and negative changes scale has been found to be 0.81. The two factors have good properties of convergent and discriminant validity and has been found to relate to posttraumatic stress and psychological distress in a consistent way.	
Curiosity and Exploration Inventory	CEI	Yes	7	7-point Likert-type scale	2 Minutes	Assessing individual differences in the recognition, pursuit, and integration of novel and challenging experiences and information. The CEI has good internal reliability, and shows moderately large positive relationships with intrinsic motivation, reward sensitivity, openness to experience, and subjective vitality.	1. Kashdan, T.B., Rose, P., & Fincham, F.D. (2004). Curiosity and Exploration: Facilitating positive subjective experiences and personal growth opportunities. <i>Journal of Personality Assessment</i> , 82(3), 291-305. 2. Kashdan, T.B. (2002). Social anxiety dimensions, neuroticism, and the contours of positive psychological functioning. <i>Cognitive Therapy and Research</i> , 26, 789-810. 3. Kashdan, T.B. (2004) The neglected relationship between social interaction anxiety and hedonic deficits: Differentiation from depressive symptoms. <i>Journal of Anxiety Disorders</i> , 18, 719-730.

Questionnaire Title	Questionnaire Title Abbreviation	Self-Report	Number of items	Response Format	Completion Time	Other Details	Key References
Gratitude Questionnaire – 6	GQ-6	Yes	6	7-point Likert-type scale	5 Minutes	GQ-6 measures the disposition to experience gratitude. It has good internal reliability, with alphas between .82 and .87, and there is evidence that the GQ-6 is positively related to optimism, life satisfaction, hope, spirituality and religiousness, forgiveness, empathy and prosocial behaviour, and negatively related to depression, anxiety, materialism and envy.	1. McCullough, M. E., Emmons, R. A., & Tsang, J. (2002). The Grateful Disposition: A conceptual and Empirical Topography. <i>Journal of Personality and Social Psychology</i> , 82, 112-127.
Inspiration Scale	IS	Yes	8	7-point Likert-type scale	2 Minutes	IS demonstrates a consistent two-factor structure, internal consistency, temporal stability, and measurement invariance across time and across populations. It has construct validity.	1. Thrash, T. M., & Elliot, A. J. (2003). Inspiration as a psychological construct. <i>Journal of Personality and Social Psychology</i> , 84, 871-889.
Meaning in Life Questionnaire	MLQ	Yes	10	7-point Likert-type scale	5 Minutes	MLQ has good reliability, test-retest stability, stable factor structure, and convergence among informants. Presence is positively related to well-being, intrinsic religiosity, extraversion and agreeableness, and negatively related to anxiety and depression. Search is positively related to religious quest, rumination, past-negative and present-fatalistic time perspectives, negative affect, depression, and neuroticism, and negatively related to future time perspective, close-mindedness (dogmatism), and well-being. Presence relates as expected with personal growth self-appraisals, and altruistic and spiritual behaviours as assessed through daily diaries.	1. Steger, M. F., Frazier, P., Oishi, S., & Kaler, M. (2006). The Meaning in Life Questionnaire: Assessing the presence of and search for meaning in life. <i>Journal of Counseling Psychology</i> , 53(1), 80-93. 2. Steger, M. F., & Frazier, P. (2005). Meaning in life: One link in the chain from religion to well-being. <i>Journal of Counseling Psychology</i> , 52(4), 574-582.

Questionnaire Title	Questionnaire Title Abbreviation	Self-Report	Number of items	Response Format	Completion Time	Other Details	Key References
Mindful Attention Awareness Scale	MAAS	Yes	15	6-point Likert-type scale	10 Minutes	MAAS is designed to assess a core characteristic of dispositional mindfulness, namely, open or receptive awareness of and attention to what is taking place in the present. The scale shows strong psychometric properties.	1. Brown, K.W. & Ryan, R.M. (2003). The benefits of being present: Mindfulness and its role in psychological well-being. <i>Journal of Personality and Social Psychology</i> , 84, 822-848.
Older Adults' Attributional Style Questionnaire	OAASQ	Yes	12	7-point Likert-type scale	20 Minutes	OAASQ is a version of the ASQ modified to be appropriate to the lives of older adults.	1. Isaacowitz, D.M. & Seligman, M.E.P. (2002). Cognitive style predictors of affect change in older adults. <i>International Journal of Aging and Human Development</i> , 54(3), 233-253. 2. Isaacowitz, D.M. & Seligman, M.E.P. (2001). Is pessimistic explanatory style a risk factor for depressive mood among community-dwelling older adults? <i>Behaviour Research and Therapy</i> , 39, 255-272.
Perceived Benefits Scale	PBS	Yes	38	5-point Likert-type scale	Not stated	Cronbach's alpha reliability coefficients for the PBS positive scales range from 0.73 to 0.93, and test-retest correlation coefficient reported over 2 weeks range from 0.66 to 0.97. Information for the PBS negative scale was not reported. Strong correlations between PBS and PTGI were reported.	1. McMillen, J.C., & Fisher, R.H. (1998) The Perceived Benefits Scales: Measuring perceived positive life changes after negative events. <i>Social Work Research</i> , 22(3), 173-186.
Personal Growth Initiative Scale	PGIS	Yes	9	6-point Likert-type scale	5 Minutes	PGIS is scale for personal growth initiative. Personal growth initiative is a person's active and intentional involvement in changing and developing as a person. There is evidence that the PGIS is strongly positively related to psychological well-being and negatively related to psychological distress. Reliability and validity evidence has been strong.	1. Bartley, D. F., & Robitschek, C. (2000). Career exploration: A multivariate analysis of predictors. <i>Journal of Vocational Behavior</i> , 56, 63-81. 2. Robitschek, C. (1998). Personal growth initiative: The construct and its measure. <i>Measurement and Evaluation in Counseling and Development</i> , 30, 183-198. 3. Robitschek, C. (1999). Further validation of the Personal Growth Initiative Scale. <i>Measurement and Evaluation in Counselling and Development</i> , 31, 197-210. 4. Robitschek, C., & Cook, S. W. (1999). The influence of personal growth initiative and coping styles on career

Questionnaire Title	Questionnaire Title Abbreviation	Self-Report	Number of items	Response Format	Completion Time	Other Details	Key References
							exploration and vocational identity. <i>Journal of Vocational Behavior</i> , 54, 127-141. 5. Robitschek, C., & Kashubeck, S. (1999). A structural model of parental alcoholism, family functioning, and psychological health: The mediating effects of hardiness and personal growth orientation. <i>Journal of Counseling Psychology</i> , 46, 159-172. 6. Whittaker, A. E., & Robitschek, C. (2001). Multidimensional family functioning as predictors of personal growth initiative. <i>Journal of Counseling Psychology</i> , 48, 420-427.
Post Traumatic Growth Inventory	PTGI	Yes	21	7-point Likert-type scale	Not stated	Internal consistency coefficients ranged from 0.67 to 0.85 for the subscales. The alpha coefficient for the normative sample was 0.90. Test-retest reliability measured two months later, was 0.71 for the total score, but 0.37 for some of the sub-scales.	1. Tedeschi, R.G., & Calhoun, L.G. (1996) Posttraumatic Growth Inventory: Measuring the positive latency of trauma. <i>Journal of Traumatic Stress</i> , 9, 455-471.
Psychological Well-Being Scales	PWB	Yes	42	6-point Likert-type scale	Not stated	This scale conceptualises psychological well-being as consisting of 6 dimensions: autonomy, environmental mastery, personal growth, positive relations with others, purpose in life, self-acceptance.	1. Ryff, C.D., & Singer, B. (1998). The contours of positive human health. <i>Psychological Inquiry</i> , 9, 1-28. 2. Ryff, C.D. (1995). Psychological well-being in adult life. <i>Current Directions in Psychological Science</i> , 4, 99-104. 3. Ryff, C.D., & Keyes, C.L.M. (1995). The structure of psychological well-being revisited. <i>Journal of Personality and Social Psychology</i> , 69, 719-727.
Quality of Life Inventory	QOLI	Yes	16	6-point Likert-type scale	Not stated	QOLI assesses an individual's quality of life through the importance they attach to each of 16 life domains, as well as their current satisfaction with each domain.	1. Frisch, M.B. (1992). Use of the Quality of Life Inventory in problem assessment and treatment planning for cognitive therapy of depression. In A. Freeman & F.M. Dattilio (Eds). <i>Comprehensive Casebook of Cognitive Therapy</i> (pp. 27-52). New York: Plenum. 2. Frisch, M.B., Cornell, J., Villanueva, M., & Retzlaff, P.J. (1992). Clinical validation of the Quality of Life Inventory: A measure of life satisfaction for use in treatment planning

Questionnaire Title	Questionnaire Title Abbreviation	Self-Report	Number of items	Response Format	Completion Time	Other Details	Key References
							and outcome assessment. Psychological Assessment: A Journal of Consulting and Clinical Psychology, 4, 92-101.
Satisfaction with Life Scale	-	Yes	5	7-point Likert-type scale	Few Minutes	The Satisfaction with Life Scale was developed to assess satisfaction with people's lives as a whole. The scale does not assess satisfaction with specific life domains, such as health or finances, but allows subjects to integrate and weigh these domains in whatever way they choose.	1. Diener, E., Emmons, R.A., Larson, R.J., & Griffin, S. (1985). The satisfaction with life scale. Journal of Personality Assessment, 49, 71-75. 2. Pavot, W. & Diener, E. (1993). Review of the Satisfaction with Life Scale. Psychological Assessment, 5, 164-172. 3. Pavot, W. G., Diener, E., Colvin, C. R., & Sandvik, E. (1991). Further validation of the Satisfaction with Life Scale: Evidence for the cross-method convergence of well-being measures. Journal of Personality Assessment, 57, 149-161.
Silver Lining Questionnaire	SLQ	Yes	38	5-point Likert-type scale	Not stated	The Silver Lining Questionnaire measures the extent to which people believe their illness has had a positive benefit despite the negative consequences of being ill. Research suggests that this positive interpretation is not due to a form of self-delusion but instead reflects personal growth and that it can be enhanced by the context.	1. Sodergren, S. C. & Hyland, M. E. (1997). Qualitative phase in the development of the Silver Lining Questionnaire. Quality of Life Research, 6, (7-8), 365. 2. Sodergren, S. C., & Hyland, M. E. (2000). What are the positive consequences of illness? Psychology and Health, 15, 85-97. 3. Sodergren, S. C., Hyland, M. E., Singh, S. J., & Sewell, L. (2002). The effect of rehabilitation on positive interpretations of illness. Psychology and Health; 17, 753-760. 4. Sodergren, S. C., Hyland, M. E., Crawford, A., Partridge, M. R. (2004). Positivity in illness: self-delusion or existential growth? British Journal of Health Psychology, 9, 163-174. 5. Hyland, M. E., Sodergren, S. C., & Lewith, G. T. (2006). The role of positivity in chronic fatigue syndrome (CFS). Journal of Health Psychology, 11(5), 731-741.

Questionnaire Title	Questionnaire Title Abbreviation	Self-Report	Number of items	Response Format	Completion Time	Other Details	Key References
State-Trait-Cheerfulness Inventory	STCI	Yes	20 / 10	4-point Likert-type scale	10 / 5 Minutes	STCI instrument measures the three concepts of cheerfulness, seriousness, and bad mood as both states (STCI-S) and traits (STCI-T). The concepts are considered to assess the temperamental basis of humour and the scales have been validated in a variety of studies	1. Ruch, W., Kohler, G. & van Thriel (1996). Assessing the "humorous temperament": Construction of the facet and standard trait forms of the State-Trait-Cheerfulness-Inventory - STCI. In W. Ruch (Ed.), Measurement of the sense of humor [special issue]. <i>Humor: International Journal of Humor Research</i>, 9, 303-339. 2. Ruch, W., Kohler, G. & van Thriel (1997). To be in good or bad humor: Construction of the state form of the State-Trait-Cheerfulness-Inventory - STCI. <i>Personality and Individual Differences</i>, 22, 477-491. 3. Ruch, W. & Kohler, G. (1998). A temperament approach to humor. In: W. Ruch (Ed.), <i>The sense of humor: Explorations of a personality characteristic</i>. (Humor Research Series, vol. 3). Berlin: Mouton de Gruyter, 203-230. 4. Ruch, W. & Carrell, A. (1998). Trait cheerfulness and the sense of humor. <i>Personality and Individual Differences</i>, 24, 551-558. 5. Ruch, W. (1997). State and trait cheerfulness and the induction of exhilaration. <i>European Psychologist</i>, 2, 328-341.
Stress-Related Growth Scale	SRGS	Yes	50	4 point Likert-type scale	Not stated	Internal consistency coefficients for the SRGS in the mid 0.90s. Two week test-retest reliability was 0.81.	Park, C.L., Cohen, L.H., & Murch, R. (1996). Assessment and prediction of stress related growth. <i>Journal of Personality</i> , 64, 71-105.

Questionnaire Title	Questionnaire Title Abbreviation	Self-Report	Number of items	Response Format	Completion Time	Other Details	Key References
Subjective Happiness Scale	SHS	Yes	4	7-point Likert-type scale	Not stated	SHS is a global subjective happiness scale. SHS has high internal consistency, which has been found to be stable across samples. Test-retest and self-peer correlations have suggested good to excellent reliability, and construct validation studies of convergent and discriminant validity have confirmed the use of this scale to measure the construct of subjective happiness.	1. Lyubomirsky, S., & Lepper, H. S. (1999). A measure of subjective happiness: Preliminary reliability and construct validation. <i>Social Indicators Research</i> , 46, 137-155. 2. Lyubomirsky, S., & Ross, L. (1997). Hedonic consequences of social comparison: A contrast of happy and unhappy people. <i>Journal of Personality and Social Psychology</i> , 73, 1141-1157. 3. Lyubomirsky, S., & Ross, L. (1999). Changes in attractiveness of elected, rejected, and precluded alternatives: A comparison of happy and unhappy individuals. <i>Journal of Personality and Social Psychology</i> , 76, 988-1007. 4. Lyubomirsky, S., & Tucker, K. L. (1998). Implications of individual differences in subjective happiness for perceiving, interpreting, and thinking about life events. <i>Motivation and Emotion</i> , 22, 155-186. 5. Lyubomirsky, S. (2001). Why are some people happier than others?: The role of cognitive and motivational processes in well-being. <i>American Psychologist</i> , 56, 239-249.
Thriving Scale	TS	Yes	20	5-point Likert-type scale	Not stated	Cronbach's alpha reliability coefficient for the TS of 0.92. No test-retest information is reported.	1. Abraido-Lanza, A.F., Guier, C., & Colon, R.M. (1998). Psychological thriving among Latinas with chronic illness. <i>Journal of Social Issues</i> , 54, 405-424.
Transgression-Related Interpersonal Motivations Inventory	TRIM	Yes	12	5-point Likert-type scale	5 Minutes	TRIM assesses the motivations assumed to underlie forgiving: Avoidance and Revenge. TRIM subscales not only correlate with a variety of relationship, offense, and social-cognitive variables, they have also demonstrated strong relationships to a single-item measure of forgiveness.	1. McCullough, M. E., Rachal, K.C., Sandage, S. J., Worthington, E. L., Brown, Susan W., & Hight, T. L.(1998). Interpersonal Forgiving in Close Relationships: II. Theoretical Elaboration and Measurement. <i>Journal of Personality and Social Psychology</i> , 75, 1586-1603. 2. McCullough, M. E., Hoyt, W. T., & Rachal, K. C. (2000). What We Know (and Need to Know) about Assessing Forgiveness Constructs. In McCullough, M. E., Pargament, K.I., & Carl E. Thoresen (Eds.), <i>Forgiveness: Theory, Research, and Practice</i> (pp.65-88). New York: Guilford

Questionnaire Title	Questionnaire Title Abbreviation	Self-Report	Number of items	Response Format	Completion Time	Other Details	Key References
							Publications, Inc. 3. McCullough, M. E., Bellah, C.G., Kilpatrick, S. D., & Johnson, J. L. (2001). Vengefulness: Relationships With Forgiveness, Rumination, Well-Being, and the Big Five. <i>Journal of Personality and Social Psychology Bulletin</i> , 27, 601-610.
VIA Inventory of Strengths	VIA-IS	Yes	240	5-point Likert-type scale	25 Minutes	VIA-IS has face-validity and is intended for use with adults in the contemporary United States.	1. Peterson, C., & Seligman, M. E. P. (in preparation). The VIA classification of strengths. Cincinnati, OH: Values in Action Institute. 2. Peterson, C., & Seligman, M. E. P., (2001). VIA Inventory of Strengths (VIA-IS).

Appendix 17: Impact of research of paper 7

Systematic reviews do not easily lend themselves to conference presentations (oral or poster), so my focus for the dissemination of this work, was also a peer review journal publication. Due to the growing popularity of PPC, and the limited number of publications within HNC I was very keen to publish this review in an open access journal. At the time, I was only really aware of PeerJ and PLOS ONE, and also that there were a growing body of 'journals' that were *pay to publish*. These later types were not indexed on the traditional databases (Scopus, AIDS Abstracts, SIIC Data Bases, Current Contents/Life Sciences and Clinical Medicine, MEDLINE®, Excerpta Medica, Science Citation Index, Current Awareness in Biological Sciences, EMBASE), so once published no one would easily find the review to reference. At the time I choose PeerJ it had not been published for long enough to have an impact factor, but a year later it was rated as 2.1 and had a Source Normalized Impact per Paper value of 0.84.

At the time of writing and excluding self-citations, Mendeley indicated that it has been cited **19** times, all of which can be identified through the Web of Science services:

1. Zhang MM, Yang YJ, Su D, Zhang T, Jiang XX & Li HP. (2019) [A randomized controlled trial of a guided self-disclosure intervention to facilitate benefit finding in Chinese breast cancer patients: study protocol](#). Journal of Advanced Nursing, doi: 10.1111/jan.14042.
2. Dunne S, Coffey L, Sharp L, Desmond D, Cullen C, O'Connor J, O'Sullivan E, Timon C & Gallagher P. (2019) [Investigating the impact of self-management behaviours on quality of life and fear of recurrence in head and neck cancer survivors: A population-based survey](#). Psycho-Oncology, 28(4):742-749. doi10.1002/pon.5010.
3. Leong Abdullah MFI, Hami R, Appalanaido GK, Azman N, Shariff NM, Md Sharif SS. (2019) [Diagnosis of cancer is not a death sentence: Examining posttraumatic growth and its associated factors in cancer patients](#). Journal of Psychosocial Oncology Mar 1:1-16. doi: 10.1080/07347332.2019.1574946.
4. Wu X, Kaminga AC, Dai W, Deng J, Wang Z, Pan X & Liu A. (2019)[The prevalence of moderate-to-high posttraumatic growth: A systematic review](#)

- [and meta-analysis](#). Journal of Affective Disorders. 15;243:408-415. doi: 10.1016/j.jad.2018.09.023.
5. Lopes CB, Magalhães LL, Teófilo CR, Alves APNN, Montenegro RC, Negri M & Ribeiro-dos-Santos A. (2018) [Differential expression of hsa-miR-221, hsa-miR-21, hsa-miR-135b, and hsa-miR-29c suggests a field effect in oral cancer](#) BMC Cancer. 6;18(1):721. doi: 10.1186/s12885-018-4631-z.
 6. Li Q, Lin L, Zhou H, Xu Y, Yang L, & Xu Y. (2018) [Factors moderating the mutual impact of benefit finding between Chinese patients with cancer and their family caregivers: A cross-sectional study](#) Psycho-Oncology. 27(10):2363-2373. doi: 10.1002/pon.4833.
 7. Shand LK, Brooker JE, Burney S, Fletcher J & Ricciardelli LA. (2018) [Psychosocial factors associated with posttraumatic stress and growth in Australian women with ovarian cancer](#) Journal of Psychosocial Oncology. 36(4):470-483. doi: 10.1080/07347332.2018.1461728.
 8. Olson A, Reiland S, Davies S & Koehler AR. (2018) [Learning about the experience of living with chronic conditions: A framework analysis of nursing students' reflections on their conversations with older adults](#). Gerontology & Geriatrics Education. 39(3):295-315. doi: 10.1080/02701960.2016.1247067.
 9. Kendell K & Armstrong NJ. (2018) [Survivorship: The Role of the Clinical Psychologist and the Clinical Nurse Specialist in Thyroid Cancer Care](#). Practical Management of Thyroid Cancer, Chapter 27 pp 369-385
 10. Litzelman K, Blanch-Hartigan D, Lin CC & Han X. (2017) [Correlates of the positive psychological byproducts of cancer: Role of family caregivers and informational support](#) Palliative & Supportive Care. 15(6):693-703. doi: 10.1017/S1478951517000050.
 11. Jones SMW, Ziebell R, Walker R, Nekhlyudov L, Rabin BA, Nutt S, Fujii M & Chubak J. (2017) [Association of worry about cancer to benefit finding and functioning in long-term cancer survivors](#). Supportive Care in Cancer. 25(5):1417-1422. doi: 10.1007/s00520-016-3537-z.
 12. Gonzalez BD, Manne SL, Stapleton J, Myers-Virtue S, Ozga M, Kissane D, Heckman C & Morgan M. (2017) [Quality of life trajectories after diagnosis of](#)

- [gynecologic cancer: a theoretically based approach](#). Supportive Care in Cancer. 25(2):589-598. doi: 10.1007/s00520-016-3443-4.
13. Holtmaat K, van der Spek N, Cuijpers P, Leemans CR & Verdonck-de Leeuw IM. (2017) [Posttraumatic growth among head and neck cancer survivors with psychological distress](#). Psycho-Oncology, 26(1):96-101. doi: 10.1002/pon.4106.
 14. Ruini C. (2017) [Positive Psychology in the Clinical Domains](#). Research and Practice, Dordrecht, Springer Netherlands, 2017, pp. 206
 15. Keitel MA & Wertz LH. (2017) [Gender and Meaning Making. The Experiences of Individuals With Cancer](#), Reconstructing Meaning After Trauma, Chapter 4, pp 47-68
 16. Adams SV, Ceballos R & Newcomb PA. (2016) [Quality of Life and Mortality of Long-Term Colorectal Cancer Survivors in the Seattle Colorectal Cancer Family Registry](#). PLoS ONE , 2;11(6):e0156534. doi: 10.1371/journal.pone.0156534.
 17. Ridgway E, Grose J, Charles A, Hewett J, Jarvis M & Benjamin S. (2016) [The experience of patients with a 'good' cancer](#). European Journal of Cancer Care. 25(3):348-55. doi: 10.1111/ecc.12258.
 18. Chiba R, Miyamoto Y & Harada N, (2016) [Psychological Transformation by an Intervention to Facilitate Benefit Finding Among People With Chronic Mental Illness in Japan](#). Perspectives In Psychiatric Care. 52(2):139-44. doi: 10.1111/ppc.12110.
 19. Martz E & Livneh H. (2016) [Psychosocial Adaptation to Disability Within the Context of Positive Psychology: Findings from the Literature](#). Journal of Occupational Rehabilitation. 26(1):4-12. doi: 10.1007/s10926-015-9598-x.

Paper 7, at the time of writing, has a Mendeley statistic of 43.

As with the papers in previous chapters, Dr Phil Clatworthy and I applied a QA tool. Once again we used the Joanna Briggs Institutes range of tools and for paper 3 we used the Case Series checklist.

Quality Appraisal - Systematic Reviews (Aromatataris, Fernandez, Godfrey et al 2015)

	Yes	No	Unclear	Not applicable
1. Is the review question clearly and explicitly stated?	☒	☐	☐	☐
2. Were the inclusion criteria appropriate for the review question?	☒	☐	☐	☐
3. Was the search strategy appropriate?	☒	☐	☐	☐
4. Were the sources and resources used to search for studies adequate?	☒	☐	☐	☐
5. Were the criteria for appraising studies appropriate?	☒	☐	☐	☐
6. Was critical appraisal conducted by two or more reviewers independently?	☒	☐	☐	☐
7. Were there methods to minimize errors in data extraction?	☐	☐	☒	☐
8. Were the methods used to combine studies appropriate?	☒	☐	☐	☐
9. Was the likelihood of publication bias assessed?	☐	☒	☐	☐
10. Were recommendations for policy and/or practice supported by the reported data?	☐	☒	☐	☐
11. Were the specific directives for new research appropriate?	☒	☐	☐	☐

Appendix 18: Impact of research of paper 8

As mentioned above systematic reviews do not lend themselves to conference presentations, and as such Anna did not present the review as a standalone piece of work, although did refer to it in subsequent presentations of her PhD research.

The paper was published in *Communication Disorders Quarterly*, which at the time of publishing paper 8 had an impact factor of 0.5 and a Source Normalized Impact per Paper value of 0.908. The *Communication Disorders Quarterly* is an international journal aimed at speech and language pathologists/therapist and teachers of the deaf and hard of hearing. As a Sage Publishing journal, it is indexed on the usual databases (Cumulative Index to Nursing Administration and Health Literature, Contents Pages in Education (T&F), Educational Research Abstracts Online (T&F), Gale: Expanded Academic ASAP, MediaFinder, NISC, ProQuest: Linguistics and Language Behavior Abstracts (LLBA), PsycINFO, Scopus).

At the time of writing and excluding self-citations, Mendeley indicated that it has been cited **6** times, all of which can be identified through the web of science services:

- 1) McDonald D, Colmer S, Guest S, Humber D, Ward C & Young J. (2019) [Parent-implemented language intervention delivered by therapy assistants for two-year-olds at risk of language difficulties: A case series](https://doi.org/10.1177/0265659019842244). *Child Language Teaching and Therapy*. <https://doi.org/10.1177/0265659019842244>
- 2) Yuill N & Little S. (2018) [Thinking or feeling? An exploratory study of maternal scaffolding, child mental state talk, and emotion understanding in language-impaired and typically developing school-aged children](https://doi.org/10.1111/bjep.12194). *British Journal of Educational Psychology*. 88(2), 261-283, <https://doi.org/10.1111/bjep.12194>
- 3) Green KB, Towson JA, Head C, Janowski B & Smith L. (2018) [Facilitated playgroups to promote speech and language skills of young children with communication delays: A pilot study](https://doi.org/10.1177/0265659018755525) *Child Language Teaching and Therapy*. 34(1), 37-52, <https://doi.org/10.1177/0265659018755525>
- 4) Lüke C, Ritterfeld U, Grimmering A, Liszkowski U & Rohlfing KJ. (2017) [Development of Pointing Gestures in Children With Typical and Delayed Language Acquisition](https://doi.org/10.1044/2017_jslhr-l-16-0129). *Journal of Speech Language and Hearing Research*. 60(11), 3185-3197, https://doi.org/10.1044/2017_jslhr-l-16-0129

- 5) Zevenbergen AA, Holmes A, Haman E, Whiteford N & Thielges S. (2016) [Variability in mothers' support for preschoolers' contributions to co-constructed narratives as a function of child age](https://doi.org/10.1177/0142723716673955). First Language - 36(6), 601-616, <https://doi.org/10.1177/0142723716673955>
- 6) Medeiros KF & Cress CJ. (2016) [Differences in maternal responsive and directive behavior during free play with and without aided AAC](https://doi.org/10.1080/07434618.2016.1179341). Augmentative and Alternative Communication. 32(2), 151-161, <https://doi.org/10.1080/07434618.2016.1179341>

Paper 8, at the time of writing, has a Mendeley statistic of 46.

As with the papers in previous chapters, Dr Phil Clatworthy and I applied a QA tool. Once again we used the Joanna Briggs Institutes range of tools and for paper 3 we used the Case Series checklist.

Quality Appraisal - Systematic Reviews (Aromatataris, Fernandez, Godfrey et al 2015)

	Yes	No	Unclear	Not applicable
1. Is the review question clearly and explicitly stated?	☒	☐	☐	☐
2. Were the inclusion criteria appropriate for the review question?	☒	☐	☐	☐
3. Was the search strategy appropriate?	☒	☐	☐	☐
4. Were the sources and resources used to search for studies adequate?	☒	☐	☐	☐
5. Were the criteria for appraising studies appropriate?	☒	☐	☐	☐
6. Was critical appraisal conducted by two or more reviewers independently?	☒	☐	☐	☐
7. Were there methods to minimize errors in data extraction?	☐	☒	☐	☐
8. Were the methods used to combine studies appropriate?	☒	☐	☐	☐
9. Was the likelihood of publication bias assessed?	☐	☒	☐	☐
10. Were recommendations for policy and/or practice supported by the reported data?	☐	☒	☐	☐
11. Were the specific directives for new research appropriate?	☒	☐	☐	☐

Appendix 19: Impact of research reported of paper 9

As with the previous two papers (7 and 8), this work was not presented prior to its dissemination via paper 9. The paper was published in the International Journal of Language and Communication Disorders. At the time of publishing paper 9, the International Journal of Language and Communication Disorders had an impact factor of 1.50 and a Source Normalized Impact per Paper value of 1.214. This journal is also indexed on a surprisingly large number of databases (Abstracts on Hygiene & Communicable Diseases (CABI), Academic Search Alumni Edition (EBSCO Publishing), British Education Index (EBSCO Publishing), CAB Abstracts® (CABI), Current Contents: Social & Behavioral Sciences (Clarivate Analytics), ERIC: Educational Resources Information Center (CSC), Global Health (CABI), Health & Medical Collection (ProQuest), Health Research Premium Collection (ProQuest), Hospital Premium Collection (ProQuest), Linguistics Collection (ProQuest), MLA International Bibliography (MLA), ProQuest Central (ProQuest), ProQuest Central K-253, ProQuest Central K-254, Psychology Database (ProQuest), Science Citation Index Expanded (Clarivate Analytics), Social Science Premium Collection (ProQuest), Social Sciences Citation Index (Clarivate Analytics), Web of Science (Clarivate Analytics)), and is picked up multiple time when running the type of searches used during systematic reviewing.

At the time of writing, Mendeley indicates that it has been cited **3** times, all of which can be identified through Web of Science services:

1. Alsaad M, McCabe P & Purcell A. (2019) The Application of the Maximal Opposition Therapy Approach to an Arabic-Speaking Child, *Journal of Communication Disorders*, 10.1016/j.jcomdis.2019.105913, (105913).
2. Hegarty N, Titterton J, McLeod S & Taggart L (2018) Intervention for children with phonological impairment: Knowledge, practices and intervention intensity in the UK, *International Journal of Language & Communication Disorders*, **53**, 5, (995-1006)
3. Nakamichi N, Takamoto K, Nishimaru H, Fujiwara K, Takamura Y, Matsumoto J, Noguchi M & Nishijo H. (2018) Cerebral Hemodynamics in Speech-Related Cortical Areas: Articulation Learning Involves the Inferior Frontal Gyrus, Ventral Sensory-Motor Cortex, and Parietal-Temporal Sylvian Area, *Frontiers in Neurology*, 10.3389/fneur.2018.00939, **9**.

Paper 9, at the time of writing, has a Mendelet statistic of 53. The paper has yet to be cited many times, probably due to only being published at the start of 2018. This is likely due to the lag between publishing, reading and informing fellow researchers' thinking. However, the paper was commended as being one of the top 20 articles from the International Journal of Language and Communication Disorders downloaded in 2018 (Appendix 21), so we hope it will be cited a lot in the near future.

As with the papers in previous chapters, Dr Phil Clatworthy and I applied a QA tool. Once again we used the Joanna Briggs Institutes range of tools and for paper 9 we used the Systematic Review checklist.

Quality Appraisal - Systematic Reviews (Aromatataris, Fernandez, Godfrey et al 2015)

	Yes	No	Unclear	Not applicable
1. Is the review question clearly and explicitly stated?	☒	☐	☐	☐
2. Were the inclusion criteria appropriate for the review question?	☒	☐	☐	☐
3. Was the search strategy appropriate?	☒	☐	☐	☐
4. Were the sources and resources used to search for studies adequate?	☒	☐	☐	☐
5. Were the criteria for appraising studies appropriate?	☒	☐	☐	☐
6. Was critical appraisal conducted by two or more reviewers independently?	☒	☐	☐	☐
7. Were there methods to minimize errors in data extraction?	☐	☐	☒	☐
8. Were the methods used to combine studies appropriate?	☒	☐	☐	☐
9. Was the likelihood of publication bias assessed?	☐	☒	☐	☐
10. Were recommendations for policy and/or practice supported by the reported data?	☒	☐	☐	☐
11. Were the specific directives for new research appropriate?	☒	☐	☐	☐

Appendix 20: Certificate of recognition



Appendix 21: Southmead Hospital Charity, Expression of Interest – Male Breast Cancer

1. Project Details	
Project title (To be written in plain English):	Men have breasts too - What is it like to be a man with breast cancer?
Start date:	1st March 2020
Project duration (months):	12 months
Total funding requested (estimated):	

2. Lead Applicant Details	
Name:	Sam Harding
Department:	Research and Innovation
Job Title:	Senior Research Associate
Email (for all correspondence):	Samantha.Harding@nbt.nhs.uk
Telephone:	43957
NBT contract held:	Substantive
If Honorary, where is your main contract held?	
Qualifications held:	DHealthPSych, MPhil, MSc, BSc

3. Co-Applicant Details					
Surname	First Name(s)	Job Title	Institution and/or Department	Email	Telephone
Jones	Lyn	Radiologist	Breast Care	Lyn.Jones@nbt.nhs.uk	0117 4149016
Dunn	Janet	Deputy Director of Warwick Clinical Trials Unit	University of Warwick	j.a.dunn@warwick.ac.uk	-
McCleane	Stuart	Health and Wellbeing lead, Centre for Public Health and Wellbeing	University of the West of England	Stuart.Mcclean@uwe.ac.uk	011732 88783
McIntosh	James	Consultant	Royal United	jamiemcintosh@nhs.net	-

		Oncoplasti c Breast Surgeon	Hospitals NHS Foundatio n Trust		
--	--	-----------------------------------	--	--	--

(Please add more rows if applicable)

4. Scientific Summary

i. Background

Detail the size and nature of the problem to be addressed; include a brief literature review (500 word max)

Male Breast Cancer is rare, accounting for less than 1% of all breast cancer cases and 1% of cancer cases in men.¹ This means that there is little awareness among men, and even among physicians, regarding the occurrence of breast cancer in males. A major implication of this being late diagnosis and subsequent negative impact on clinical prognosis and psychological well-being.² Across all cancer sites people are now twice as likely to survive at least 10 years than they were at the start of the 1970s. Better screening and advances in treatment over the last forty years mean we have seen a huge change in what a cancer diagnosis means in terms of mortality. This has also led to changes in morbidities and treatment side effects people have to live with.⁴

Recent psychological research has focused on the potential for people diagnosed with cancer to have substantial "positive psychological change resulting from the struggle to overcome highly challenging life circumstances".⁵⁻¹¹ In some cases these benefits merely mitigate the negative consequences of illness, but there are also instances where people report an overall benefit from being ill. These changes may concern alterations in the perceptions of oneself, social relationships with family and friends, life priorities, and appreciation of life. Cancer survivors from tumours in a range of locations frequently report having altered priorities and psychology,^{7,12-15} but the pattern and time course of these changes has been found to differ by age, location, tumour stage, and gender.

It is becoming clear that both good and bad can come from the diagnosis and treatment of breast cancer, at least for women. The researchers have yet to identify breast cancer studies focusing on psychological factors that are relevant to men. Published studies investigating breast cancer most frequently have no male participants at all and in the rare cases that they do, they do not reach a representative number of men e.g. approximately 1 in 100. Clinicians are therefore reliant on their own patients to understand men's perspectives, but that this is hampered by the low numbers any one clinician will see.

ii. Aim(s) and Objectives

Detail the research question and how this is going to be addressed (200 word max)

Research Question:

What are the psychological experiences of men who have been treated for breast cancer?

Aim:

The aim of the proposed study is to identify the lived experiences of men who have been treated for breast cancer and how these compare to literature on women's experiences. This will inform a future project to develop bespoke materials for male patients and staff working with them.

Objectives:

1. A systematic review of the literature
2. Undertake in-depth interviews (using interpretive phenomenology) with 5 men who have been treated for breast cancer in the last 5 years
3. Engage with social media platforms to form a group of men who have experienced breast cancer, who will be willing to work with the Chief Investigator to develop future grants.

iii. Plan of investigation and methodology

Briefly include all stages of the study design. Methods of data collection, measures and techniques of analysis should be described and justified for both qualitative and quantitative designs (500 word max)

Methodology:

1. Systematic Review of relevant literature following PRISMA guidelines and formulated using PICO. Steps to complete this review will include:

- 1) Define research question and inclusion/exclusion criteria
- 2) Identification of search terms and search engines
- 3) Retrieve studies from search engines (Qualitative and Quantitative)
- 4) Refine retrieved references, using the inclusion/exclusion criteria
- 5) Extract data from retained studies
- 6) Quality appraise retained studies using an appropriate tool such as Critical Appraisal Skills Program
- 7) Synthesis data from retained paper
- 8) Findings will be written up for publication in a peer review journal

2. Qualitative Interviews

- 1) Ethics approval will be obtained prior to undertaking the qualitative interviews
- 2) Potential participants will be sent a letter from their consultant alerting them to the study and asking them to contact the CI if they are interested in being part of the study
- 3) It is anticipated that in-depth, semi-structured interviews will be the primary data collection methodology. These will be undertaken face-to-face or via telephone depending on what is preferred by the participant
- 4) Participants will also be asked if they have any diaries, photo's, letters or other materials that tell aspects of their journey through and beyond their cancer treatment, that they would be willing to share with the CI
- 5) Interpretative Phenomenological Analysis (IPA) will be used. IPA is a qualitative approach developed within psychology for the examination of personal lived experience. IPA is concerned with examining lived experience, as far as possible, in its own terms as opposed to being overly influenced by prior psychological theorising or researcher bias. IPA does recognise that the exploration of the meaning of personal experience is an

interpretative endeavour on the part of both participant and researcher. This methodology will ensure that the individual lived experience is reflected rather than simply described through existing frameworks established primarily with female breast cancer patients.

6) Narrative findings will be written up for publication in a peer review journal

3. Social media - Patient Public Involvement

1) Participants from the qualitative interviews will be asked if they would be willing/interested in being part of the PPI group for this and future work. They will also be asked to review current literature, such as information sheets, to gain understanding of how relevant they are for men and how they could be amended to reflect a man's journey through breast cancer

2) The CI will undertake a programme of work to identify and make contact with established male breast cancer groups. The nature of social media will mean that these interactions are likely to transcend national boundaries. The aim is to establish a relationship with these groups and either work with each organisation to form PPI working groups, or to form an independent group from men across the existing groups. Examples of these are:

<https://www.facebook.com/MaleBreastCancerCoalition/>

<https://www.facebook.com/malebreastcancerawareness47/>

Twitter - @MaleBreastStudy

Twitter - @MBCC_MHBT

<https://plus.google.com/u/0/+TimeslikethesefilmOrg>

5. Relevance to the NHS and Potential Impact

Detail how your research project is relevant to the NHS and its potential impact e.g. potential patient benefit, service improvement, cost savings, generalisability. Explain how the gaps in knowledge will be addressed by your project and why it is important to carry out the research now (300 words max)

Male breast cancer is a rare and under-studied disease. As such, it is not as immediately attractive to funders as generic breast cancer. This has led to the position where men experience the same excellent care as women, but psychologically this may not be as suitable or beneficial for them. Men may have different needs that at present are not addressed either by psychological services or more globally by the multi-disciplinary team.

The proposed work will be the first to synthesise and evaluate psychosocial research with men with breast cancer, providing an understanding of the current state of the literature. The interview study will provide lived experiences that will enhance the literature and highlight areas of practice within breast cancer that can be amended to better treat men.

The work will have direct patient benefit at NBT. Findings will be disseminated locally, allowing all teams in the cancer pathway to understand the challenges male breast cancer patients face, which differ from their female patients.

The insights into the experiences of being a man with breast cancer will enable psychological services to better help coping, adjustment, and subsequently reducing factors identified as negative by men. Findings may be generalizable to appearance and masculinity research, and to the understanding of treatment of other cancers

such as prostate, where lived experience for men is also poorly understood. The PPI panel will advise the CI regarding the validity of the research and interpretation of the findings. This will ensure future grants are relevant to the population and produce data and materials that will directly benefit this small but significantly under-supported group. The local PPI members will guide the development of literature for use within NBT, allowing the organisation to lead the way in developing support specifically tailored to men diagnosed and being treated for breast cancer.

6. Future Direction of Research

Outline the end goal of your research project and how findings from this study will feed into future research development or NBT-led research grant applications (e.g. NIHR) or other outputs. Detail how you plan to disseminate results of your research amongst peers, patients and decision makers (300 words max)

Future research will involve:

The findings from this research will be used to inform and develop a national study. It is anticipated that this national study will develop the qualitative elements from within the current study. The qualitative work from the current study will be used to inform the content of a quantitative prospective longitudinal cross-sequential study design. The aim of which would be to gain insight into the journey of man following a diagnosis of breast cancer over at least a 5 year period.

7. Plain English Summary

This summary will be used as a stand-alone piece to judge this entire application and so please use this section to summarise your project in full, in plain English (500 words max)

Men have breasts too!

Very little is known about how men experience breast cancer, what is important to them, and how this is different from what we know from research undertaken with women.

The planned work has 3 parts:

- 1) Review research already published
- 2) Talk in depth to a number of men who have had breast cancer. Find out what it was like for them, what they thought and felt during their diagnosis, treatment, and recovery
- 3) Contact and talk with a large number of men who are members of a number of male breast cancer support groups

8. References

Please provide any references that have been used in your literature review

1. Cancer Research UK. 2016. Breast Cancer Statistics.
<http://www.cancerresearchuk.org/health-professional/cancer-statistics/statistics-by-cancer-type/breast-cancer> [Last Accessed 04/08/2016]
2. Foerster, R., Foerster, F.G., Wulff, V., Schubotz, B., Baaske, D., Wolfgarten, M.,

Kuhn, W.C., Rudlowski, C. 2011. Matched-pair analysis of patients with female and male breast cancer: a comparative analysis. *BMC Cancer*, 11:355. DOI: 1471-2407/11/335

3. Quaresma, M., Coleman, M.P., Rachet, B. 2014. *40 year trends in an index of survival for all cancers combined and survival adjusted for age and sex for each cancer in England and Wales, 1971-2011: a population-based study*. The Lancet 2014; 385: 1206-1218.
4. **Macmillan Cancer Support, 2016. Cancer: Then and Now: Diagnosis, treatment and aftercare from 1970-2016.** Research commissioned by Macmillan Cancer Support
5. Tedeschi, R. G., & Calhoun, L. G. 2004. Posttraumatic growth: Conceptual foundations and empirical evidence. *Psychological Inquiry*, 15(1), 1-18. doi:10.1207/s15327965pli1501_01
6. Collins, R.L., Taylor, S.E., Skokan, L.A., 1990. A Better World or a Shattered Vision? Changes in Life Perspectives Following Victimization. *Soc. Cogn.* 8, 263–285. doi:10.1521/soco.1990.8.3.263
7. Giovinco, G., McDougald, J., 1994. Logotherapy: A journey into meaning for people with AIDS. *Int. Forum Logother.* 17, 76–81.
8. Laerum, E., Johnsen, N., Smith, P., Larsen, S., 1988. Myocardial infarction may induce positive changes in life-style and in the quality of life. *Scand. J. Prim. Health Care* 6, 67–71.
9. LaFortune-Fredette, S.L., 1995. Breast cancer survivors: concerns and coping. *Cancer Nurs.* 18, 35–46.
10. O'Connor, A.P., Wicker, C.A., Germino, B.B., 1990. Understanding the cancer patient's search for meaning. *Cancer Nurs.* 13, 167–175.
11. Taylor, S.E., 1983. Adjustment to threatening events. A theory of cognitive adaptation. *Am. Psychol.* 38, 1161–1173.
12. Dow, K.H., Ferrell, B.R., Haberman, M.R., Eaton, L., 1999. The meaning of quality of life in cancer survivorship. *Oncol. Nurs. Forum* 26, 519–528.
13. Ferrell, B.R., Grant, M., Funk, B., Otis-Green, S., Garcia, N., 1998. Quality of life in breast cancer. Part II: Psychological and spiritual well-being. *Cancer Nurs.* 21, 1–9.
14. Pelusi, J., 1997. The lived experience of surviving breast cancer. *Oncol. Nurs. Forum* 24, 1343–1353.
15. Schroevers, M.J., Ranchor, A.V., Sanderman, R., 2004. The role of age at the onset of cancer in relation to survivors' long-term adjustment: a controlled comparison over an eight-year period. *Psycho-oncology.* 13, 740–752. doi:10.1002/pon.780

9. Health Categories (please mark all that apply)			
Blood		Metabolic and Endocrine	
Cancer	x	Musculoskeletal	
Cardiovascular		Neurological	
Oral and Gastrointestinal		Renal and Urogenital	
Congenital Disorders		Reproductive health and	

		Childbirth	
Ear		Respiratory	
Eye		Skin	
Infection		Stroke	
Inflammatory and Immune System		Generic health Relevance	
Injuries and Accidents		<u>Other (please specify)</u>	
Mental Health	x		

Appendix 22: Drawing as a novel methodology – Harding and Bradford (2019)

Original Article

SAGE Open Medicine

Drawing: A novel approach to understanding appearance change in people following treatment for head and neck cancer

SAGE Open Medicine
Volume 7: 1–10
© The Author(s) 2019
Article reuse guidelines:
sagepub.com/journals-permissions
DOI: 10.1177/2050312118820345
journals.sagepub.com/home/smo


Sam Harding¹  and John Bradford²

Abstract

Objectives: Appearance is a factor within head and neck cancer health-related quality of life measures; however, the issue of self-perceived appearance has received scant attention. Self-portraiture may provide insight into the patient's perspective, allowing people to provide viewpoints that are not easily accessible. This research investigates the methodology of drawing and responses from patient-completed questionnaires in a head and neck cancer population.

Methods: A sample of 30 people at least 3 months post-treatment for head and neck cancer were recruited. Participants completed the Derriford Appearance Scale, University of Washington Quality of Life scale, and two drawings: (1) how they recall themselves pre-treatment and (2) how they see themselves post-treatment. They were asked to discuss the methodology and their experience of it.

Results: Correlations with Derriford Appearance Scale, University of Washington Quality of Life scale, and size drawings failed to find relationships between these variables. Post-treatment drawings were significantly smaller than pre-treatment. Qualitative analysis of the drawings found differences between the images. Participants related how drawing gave an opportunity to voice concerns that questionnaires and clinic appointments did not.

Conclusion: Drawing can elicit distinctly different information about a person following treatment for head and neck cancer than that provided by health-related quality of life measures. Further research would clarify if clinical opinion matches patients' drawing.

Keywords

Drawings, head and neck cancer, appearance, Derriford Appearance Scale

Date received: 7 September 2018; accepted: 22 November 2018

Introduction

It is recognised that patients can develop negative perceptions of their body after treatment for cancer.^{1,2} Loss of function and external alterations in body structure have been associated with depression that can exacerbate difficulties in communication and feelings of social rejection for people who have had treatment for head and neck cancer (HNC).³ Disfigurement is a key domain that is included in Health-Related Quality of Life (HRQoL) questionnaires specific to HNC.⁴

Even though appearance is present as a factor or domain within HNC HRQoL measures, the specific issue of the self-perceived appearance of people who have been treated for HNC has received relatively scant attention.^{5,6} Most 'body image' measures have their roots in work on weight and obesity and do not accurately assess distress and dysfunction in

relation to appearance issues faced by people living with cancer. An exception to this is the Derriford Appearance Scale (DAS24), which was developed through a collaboration between plastic/reconstructive surgeons and psychologists with both clinical and non-clinical populations,⁷ including oncology patients. In addition, there is some evidence that the appearance-specific domain of the University of Washington Quality of Life (UW QoL) Questionnaire can identify individuals with appearance concerns; however, the

¹Department of Health and Social Sciences, University of the West of England, Bristol, UK
²jbsh LLP, Plymouth, UK

Corresponding author:

Sam Harding, Department of Health and Social Sciences, University of the West of England, Bristol BS16 1QY, UK
Email: sharding.jb@gmail.com



Creative Commons Non Commercial CC BY-NC: This article is distributed under the terms of the Creative Commons Attribution-NonCommercial 4.0 License (<http://www.creativecommons.org/licenses/by-nc/4.0/>) which permits non-commercial use, reproduction and distribution of the work without further permission provided the original work is attributed as specified on the SAGE and Open Access pages (<https://us.sagepub.com/en-us/nam/open-access-at-sage>).

UW QoL has not typically been used to evaluate appearance-related adjustment specifically.^{5,8,9}

Self-portraiture, or simple drawings of own body image, may provide a valuable insight into the patient's perspective.¹⁰ It offers a way to communicate other than speech, which can allow the drawers to explore the meaning of their situation by accessing material which could be suppressed and repressed by the conscious mind.¹¹ Therefore, drawing may allow people to provide viewpoints that are not easily accessible through interviews or questionnaires.¹² The produced drawings may 'illustrate ideas in a more concrete and specific way than words'.¹³ Harrow et al.¹⁴ found that some women have mental images of their (breast) cancer, which can be accessed through verbal description and drawing. These images may embody both positive and negative beliefs about cancer, which can contribute to more meaning and informed decision-making and thus, ideally, improved outcomes. Drawing may therefore be a useful method to gain insight into people's views and experiences. This may especially be the case when they have difficulties verbally either due to functional or language barriers. It may also allow individuals to access and communicate different aspects of their concerns than traditional methods.¹⁵ Drawings have been used with children to understand their cancer experiences,¹⁶ but it is still a rarely utilised method with adults.¹⁷ However, the process required to draw could lead to a more succinct presentation of meaningful facets of the participants' experiences.

The aim of the presented research is to use the novel methodology of drawing and relate that to the traditional responses from patient-completed questionnaires in a HNC population.

Methods

Ethical approvals were obtained from the local National Health Service (NHS) regulatory body (REC reference no. 10/H0107/24). Individuals provided written consent.

Participants

As a new data collection method (drawing) for this patient cohort, an opportunistic sample of 30 people at least 3 months post-treatment for HNC was recruited during a routine follow-up visit to the maxillofacial department. Thirty was selected as approximately 20% of the total patient cohort seen in the clinic during a year and 50% of the patients seen over the 5-month recruitment time frame. All people approached agreed to participate in the study. It was also the average size of previous samples used in studies designed to explore patients' perceptions of their illness.¹⁷ Although the sample was obtained by approaching people as they attended clinic (opportunistic), this sample matched the gender split for HNC and gave a representative range of cancer stages and treatment regimens (Table 1).¹⁸

Demographic data included date of birth, sex, ethnicity, and Index of Multiple Deprivation¹⁹ calculated from

postcode at time of diagnosis, occupation, and family status (married, living with partner, living alone, and living with relative/friends). Medical data included tumour site, stage at diagnosis, date of diagnosis, treatment, and date of treatment completion. Five possible treatments or combination of treatments were represented across the responders: surgery (N=9); surgery and radiotherapy (N=5); surgery, radiotherapy, and chemotherapy (N=5); radiotherapy (N=2); and radiotherapy and chemotherapy (N=9). Four tumour locations were represented: oral cavity (N=15), oropharynx (N=7), hypopharynx (N=2), and larynx (N=6). Respondents were on average 25.79 months (range: 3–80; SD: 21.3) post-treatment.

Materials

The research used patient self-portraits as a means to elicit subjective representations in a non-verbal way. Patients were asked to produce two pencil drawings or sketches. They were provided with two sheets of A4 plain white paper and asked to do a simple pencil drawing without the use of colour. The first sheet was headed 'How I remember myself prior to treatment for head and neck cancer' and the second 'How I see myself now'. This approach makes no verbal demands, which may cause discomfort to a HNC population, and operates on an entirely subjective representation. Although interviews were not planned to be included within the current research due to the study population potentially experiencing discomfort due to restricted mouth opening and/or xerostomia as a side effect of their treatment, 10 participants requested the opportunity to speak to (S.H.) to discuss their drawings and also the experience of the methodology.

The DAS24 assesses levels of distress and dysfunction in relation to cosmetic concern. Normative data are available for both clinical and non-clinical populations.⁷ The DAS24 has been validated and demonstrated to have good psychometric properties.⁷ All 24 items contribute well to the total score, and internal consistency is high ($\alpha=0.92$) and test-retest reliability (6 months) is good ($r=0.82$).⁷ The DAS24 has also been identified as a measure that shows promise as a research tool for improving understanding of how appearance affects quality of life (QOL) in HNC patients.²⁰

The UW QoL scale is a disease-specific broad measure of HRQoL for use with people who have had HNC. It has good patient acceptability, practicality, validity, reliability, and responsiveness.²¹ The UW QoL covers 12 domains: pain, appearance, activity, recreation, swallowing, chewing, speech, shoulder function, taste, saliva, mood, and anxiety. It also has two sub-scales: physical functioning and social-emotional functioning. The UW QoL has been validated by comparison to the Karnofsky scale and Sickness Impact Profile, demonstrating an average criterion validity of 0.85.²¹ It has also been found to have internal consistency between $\alpha=0.80$ and 0.79 and Test-retest of $r=0.91$.⁸

Table 1. Psychosocial characteristics of participants.

	N	Mean	SD
Gender			
Male	24		
Female	6		
Age at diagnosis	30	58.29	9.54
Family status			
Married/living with partner	28		
Living alone	2		
Living with relatives/friends	0		
Cancer stage			
I	5		
II	7		
III	3		
IV	15		
Treatment regimen			
Surgery	9		
Surgery and radiotherapy	10		
Radio with or without chemotherapy (no surgery)	11		
Months since treatment	30	19.04	14.90
UW QoL – total	30	894.66	178.18
UW QoL – Physical Function Sub-scale	30	72.67	18.41
UW QoL – Social-Emotional Function Sub-scale	30	76.01	17.47
UW QoL – domains			
Pain	30	78.33	21.51
Appearance	30	80.00	19.03
Activity	30	74.17	20.22
Recreation	30	75.00	19.70
Swallowing	30	75.67	28.61
Chewing	30	73.33	31.44
Speech	30	84.67	18.14
Shoulder	30	83.79	27.05
Taste	30	63.67	31.89
Saliva	30	58.67	36.74
Mood	30	75.83	20.22
Anxiety	30	68.00	28.33

UW QoL: University of Washington Quality of Life.

Procedure

One week prior to the clinic, potential participants were sent a letter inviting them to participate in the research. Included was an information sheet consent form and questionnaires. After their visit with the clinician, the researcher approached them to obtain consent. If completed, the questionnaire battery was collected and the participants were given the two sheets of white A4 paper noted in the “Materials” section above. The participants were asked to ‘draw a picture of what you think you looked like before your cancer treatment and another picture of what you think you look like now’. It was made clear that the researchers were not interested in drawing ability and that a sketch was fine. Participants that had not completed the questionnaires or did not want to undertake the drawings while in the hospital were provided with a freepost return envelope. Those participants that

indicated they wanted to discuss their drawings with the researcher (S.H.) were taken into a private room and field notes taken.

To minimise observer bias, one researcher who evaluated the drawings was not present at data collection. The author undertaking the data collection was a health psychologist (S.H.) and had worked with a HNC cohort previously. While both authors were PhDs with experience in both qualitative and quantitative research methods, the second author (J.B.) had minimal experience with people who had HNC.

Statistical analysis

Data from the completed scales were entered into SPSS, version 23. Patients’ drawings were scanned and imported into National Institutes of Health (NIH) Image-J software.²² The outside perimeters of the drawn head and neck and any part

of the head and neck drawn as damaged were traced and their areas, in pixels, computed by the software, and this was entered into SPSS. The percentage of the area drawn as showing change (damaged) was calculated by dividing the damaged area by the total area of the head and neck.

Wilcoxon tests were used to investigate whether those patients whose drawing included damage differed from those who did not draw damage. Spearman's rank correlation coefficients were calculated to investigate the relationships between the percentage of the head and neck drawn as damaged.

Qualitative analysis

Each drawing was qualitatively assessed by identifying prominent features in a similar fashion to that described by Broadbent et al.²³ The authors independently evaluated each drawing and recorded their notes prior to discussion. A short discussion was held for each drawing to explore any features that might be particularly important or noteworthy, such as size of the graphic or the boldness of the line. The initial assessments were used to develop a framework group of features related to the drawings, for example, the size of drawings and facial expressions/emotions. Field notes written during and directly following each (N=10) discussion with patients were reviewed with content analysis.

Results

Wilcoxon tests on age at time of diagnosis, gender, tumour staging, treatment regimen, or responses to the questionnaires found no statistical difference between those respondents that did or did not draw any visual damage on their sketches. Table 1 shows the psychosocial characteristics of participants.

Questionnaires

Spearman's rank rho correlations were performed on the responses obtained from the participants on the DAS24 and UW QoL, including the appearance domain of UW QoL, and no relationships were found.

Drawings

In total, 27 patients returned the drawings. Figure 1 shows examples of drawing done by the participants. Of the returned data, 23 did at least two drawings; one 'How I remember myself prior to treatment' and one 'How I see myself now'. Four people simply wrote 'No Change' and did not do a second drawing. Mann-Whitney tests failed to reveal any difference on age, gender, or staging of tumour

between those that did and did not return the drawing, or those that did not complete a second drawing.

Features of drawings

View. Of the 23 people returning two drawings, 3 of them did multiple drawings for how they see themselves now. This took the format often seen in arrest photography of one facing forward and the other from the side.

Size of drawings. NIH Image-J software produces a pixel count as a proxy for area of the drawing (Table 2). Where two drawings were returned by participants, the post-treatment image was smaller than the pre-treatment picture. This was supported by statistical analysis using the Wilcoxon signed-rank test between each of the dimensions (e.g. pre horizontal length and post horizontal length), which showed significant differences between the 'Horizontal' dimension ($z = -2.581$, $P \leq 0.010$), 'Vertical' dimension ($z = -2.094$, $P \leq 0.036$), and the 'Area' ($z = -3.068$, $p \leq 0.002$), with all these dimensions getting significantly smaller following treatment.

Facial expressions/emotions. Although three respondents wrote 'no change' on their second picture and a further three showed neutral emotions unchanging between pictures, other respondents clearly depicted a change in emotion between drawings, sometimes with the addition of text to provide clear understanding. The authors independently rated the emotions shown in each drawing and then discussed their thoughts and found a 100% agreement in their interpretation of the facial expressions. Table 3 shows how drawings changed in relation to the emotion shown. One picture showed the respondent crying.

Intensity of pen strokes/shading. Where people identify scarring or a change in shape, they use thicker/heavier/shaded penmanship and often supported this highlighting with text, for example, 'scars'. Shading was also used to show areas of change such as missing teeth.

Clarifying text. Sometimes, respondents provided interesting textual information supporting the drawing or providing additional information.

One person wrote, 'Sorry, but I put the operation on the right side, and it should have been on the left side'. Where another person added explanatory text on their pictures; on the first picture 'almost always happy' and on the second 'almost always depressed'.

Not all texts were apologetic or negative. One person wrote, 'I'm hopeless at art. "FAT" 19 stone', followed by on the second picture, 'Great 10' 6lbs looking good'.



Figure 1. (Continued)



Figure 1. Patient drawings.

Most frequently, people provided short phrases often with arrows to highlight areas of change such as patchy

beard, scar, no teeth, and dimple in chin due to where scare starts.

Table 2. Pixel data as proxy measure of area.

	Gender	Photo-damaged area	Pre area drawing	Post area drawing	Post damaged area ^a	Post damaged percent	Percentage area change
1	Male	259,876	8,906,220.00	8,038,272	1062	0.01	9.75 ^b
2	Male	267,138	66,866.00	14,734	4781	32.45	77.96 ^b
3	Male	101,461	13,798.00	11,003	601	5.46	20.26 ^b
4	Male	63,310	43,153.00	32,131	6141	19.11	25.54 ^b
5	Male	82,273					
6	Male	70,237	38,687.00	31,812	6641	20.88	17.77 ^b
7	Male	93,008	42,502.00	42,502	0	0.00	0.00
8	Male		54,504.00	54,504	0	0.00	0.00
9	Male		72,423.00	39,752	3149	7.92	45.11 ^b
10	Male		5394.00	1820	0	0.00	66.26 ^b
11	Female	0	7838.00	5922	0	0.00	24.45 ^b
12	Male	74,102					
13	Female	62,295	145,109.00	145,109	26,493	18.26	0.00
14	Male	167,917	62,539.00	55,198	6837	12.39	11.74 ^b
15	Male	156,633	7430.00	22,316	2258	10.12	200.35 ^c
16	Male	0	20,215.00	20,215	202	1.00	0.00
17	Female	0	61,068.00	59,863	315	0.53	1.97 ^b
18	Female	129,840	49,747.00	53,029	13,921	26.25	6.60 ^c
19	Male	0	31,803.00	31,803	0	0.00	0.00
20	Male	21,540	38,697.00	17,491	2382	13.62	54.80 ^b
21	Male	207,229	54,420.00	24,953	421	1.69	54.15 ^b
22	Female	55,418	157,445.00	147,733	12,084	8.18	6.17 ^b
23	Female	0	46,580.00	20,962	3249	15.50	55.00 ^b
24	Male	0	43,399.00	29,592	6473	21.87	31.81 ^b
25	Male	49,847	55,135.00	63,142	12,736	20.17	14.52 ^c
26	Male	51,963	35,452.00	37,311	0	0.00	5.24 ^c
27	Male	54,066	149,313.00	143,008	38,759	27.10	4.22 ^b
28	Male	0					
29	Male	101,003	157,397.00	129,596	22,500	17.36	17.66 ^b
30	Male	0	41,143.00	28,739	495	1.72	30.15 ^b

The unit of measurement for all columns excluding those containing calculated percentages and percentage change is a 'pixel' as defined by the Image-J software.²²

^aPost area drawing-pre area drawing.

^bAn increase in drawing dimension in post-treatment drawings.

^cA decrease in drawing dimension in post-treatment drawings.

Table 3. How drawings changed in relation to the emotion shown.

First drawing emotion	Second drawing emotion	Number of respondents
Sad	Happy	1
Neutral	Neutral	3
Neutral	Sad	1
Happy	Sad	6
Happy	Neutral	3
Happy	Happy	7

Drawings and questionnaires

Spearman's rank correlation coefficients did not find any significant relationship between the size or percentage of the

area of facial damage in the drawings and any of the questionnaires totals or sub-scale scores.

Field notes

A total of 10 people spoke to the researcher about their drawings. These discussions fell into two areas: (1) interpreting the content of the drawings and (2) the experience of the methodology.

Interpreting the content. Some similarities to the clarifying text were voiced; 'I'm rubbish at drawing'. Others wanted to discuss what they had included in the picture and why. For example, one lady talked about how her first drawing (Figure 1) was 'striding out' and how this showed that she liked exercise and 'getting out and meeting with people'. The

second drawing showed how she currently felt 'meh' and did not really want to go out and meet with people. This indicated that the drawing allowed for more information beyond that of appearance to be communicated.

Participants also commented on how they liked the 'freedom to draw' what they wanted rather than it being prescriptive. Although one person did say that they 'draw what I thought you [the researcher] would be most interested in' and went on to describe the content of the post-treatment image, size of the scarring and then how they felt about the treatment and their hopes for the future and symptom improvement.

Experience of the methodology. One gentleman who had undertaken the drawing and questionnaires at home returned them during his next clinic visit. He became emotional once in the private room, saying, 'I've never been given the chance to express myself this way. I felt kinda awkward, but it meant that I ended up drawing stuff I have not been asked about and have not felt able to talk about before'. Others drew a comparison to interview studies or clinic visits and related how this exercise gave them a chance to think about things in a different way before seeing the clinical team or researcher and to include things that were important to themselves.

A couple of people commented that 'It would have been great to have been able to use colour', but that they would have to be given the pencils as it is not something they have at home.

Overall, participants said that 'drawing is fundamentally more enjoyable than ticking boxes' and that 'I enjoyed the process. Doing something a bit different opens up new ways of exploring my experience of cancer'. They also suggested that if this methodology was going to be used with everyone that it 'needs to be voluntary, because although I enjoyed it, I am sure some people would hate it' and that being given time to talk the clinician through the picture is also vital as 'they might not get it, you know what I have drawn and why it's important'.

Discussion

The aim of the reported research was to use the novel methodology of drawing and investigate if the images relate to traditional responses from patient-completed questionnaires in a HNC population. No statistical relationship was found between the questionnaire totals or the appearance domain of the UW QoL or size elements of the drawings.

Although some of the participants did not complete a second drawing, reporting no change in their appearance, no-one indicated to the researcher at the time of consenting, during the process, or on the returned forms that drawing was an invalid request and not relevant to their experience of HNC. The lack of statistical agreement and the details found in the drawings indicate that the method of data collection has a direct bearing on the information communicated by the participant.

The UW QoL scale describes important daily living dysfunction or limitations that patients complain of as part of HNC or due to its treatment effects, whereas the DAS24 provides an opportunity for the respondent to recognise self-conscious elements of appearance. It focuses on the distress and dysfunction arising from body image disturbance. The patients' visual representations revealed personal and emotional accounts of their illness experience and demonstrated potential for benefits for patients. By facilitating richer data collection, drawing has been shown to be a powerful adjunct to traditional questionnaire data. The authors' observations are consistent with Guillemin's,²⁴ who claims that, despite some disadvantages, drawings, as a research method, are a means to gain the insight into a patient's world and a source of information about many aspects of illness.

Previous studies investigating the use of drawing as a research tool have found that when face-to-face interviews supplemented data from drawings, they highlight how the interview provides an opportunity to initiate further discussions.²⁴ This is supported in the presented research through not only the inclusion of clarifying text but also the informal discussions requested by a number of participants. Care needs to be exercised where researchers interpret participants' drawings without the artists input. Although formal interviews were not conducted, general conversation was entered into if initiated by the patient. It was clear from these interactions that the participants found that the drawings function as a catalyst, which helped them reflect and articulate things that they had previously found difficult to define or discuss.

These informal observations and discussions informed the drawing analysis. It was interesting to note that while the authors agreed on the drawing analysis, evaluating without additional context did elicit further questions around meaning and interpretation and the impact a post-analysis interview would have had had it been possible with the patient group. It would be of interest in future to undertake a formal interpretive interview with patients which takes into account the potential discomfit caused by treatment side effects.

Limitations

There are a number of limitations in this work, the most notable of which is the sample size. As an exploratory study to assess the use of this methodology in a HNC cohort, 30 participants represented an attainable figure in the possible time frame. It also closely matched the median average number of 32 participants (range: 4–160) from previous drawing research.¹⁷ However, the sample in this research did not provide sufficient data to allow for statistical analysis between the questionnaires and the drawing data. A further challenge was the sampling strategy used. While the opportunistic sample did match the gender split and gave a representative range of cancer stages and treatment regimens for people that had been diagnosed with HNC,¹⁸ greater insight into a patient's

perception or understanding may have been elicited if a purposive or stratified sample had been sought. An element of the methodology which could have introduced bias into the data is the location where the drawings were undertaken. In the presented research, participants could complete the drawings in the clinical environment or at home. This could mean that respondents in a clinical setting report less negative factors in order to maintain positive relationships with the clinical team and that those undertaking the research at home spend more time considering their responses. Before undertaking future research using drawing with a HNC cohort, the authors suggest working with the patient cohort to refine the methodology and to include a more prescribed data collection protocol, informed by the patients' experiences of when, where, and how often drawing and questionnaire data should be requested. In this study, a number of participants sought out the opportunity to speak with the researcher following their completion of the questionnaires and drawing; this, to some extent, allowed the research to check their interpretation of the drawing and to reflect back the participants' means. However, a formal participant check was not undertaken at the end of the data analysis, and this would need to be addressed taking the work forward.

The way in which participants were encouraged to draw could have influenced the findings obtained. In this study, people were asked to do a simple pen or pencil drawing and not to use colour. However, as shown by Michie and Abraham,²⁵ the different elements of a study's design can affect participant behaviour. Increasing the range of drawing materials available to the drawers would provide options for expression, for example, colour may reduce the use of cross-hatching or heavily drawn line to highlight an area. Requesting a drawing of a specific area of the body can target a particular topic of research interest, while a general drawing request can be open to interpretation. The latter option allows freedom of expression but can lead to uncertainty among participants about what to draw and may add in additional variables to be interpreted within research. Although reassurance that the activity was not an assessment of drawing skill was given to the participants in the presented research, it is likely that it may be insufficient to overcome, at least some, participants' initial hesitation to draw.

Interventions which incorporate imagery are already present with the cancer setting. For example, guided imagery where participants are felt to gain an increasing sense of control over cancer-related pain and anxiety using mental images²⁶ and art therapy as a medium through which stress may be reduced²⁷ and emotions expressed.²⁸ This study shows that drawing may be a useful method for eliciting conversations during clinic visits.

This study invited a before and after drawing, done in a single sitting. Future work should consider additional time points post-treatment and drawings conducted over multiple sittings. Further work with members of the patient cohort would allow researchers and clinicians to identify which

members of the multi-disciplinary team would be best to initiate the drawing, when, to which patients, and in what context.

Previous research of drawings over time of non-clinical people has observed that structural and formal aspects of drawing size, line, and placement are less subject to variability than content, such as body details, clothing, and accessories.²⁹ Whether an individual makes their figure large or small, where they place it on the page, what the essential proportions of the figure are, whether symmetry is observed, or shading is used are all features that have been shown to be stable in the non-clinical population.²⁹ It is especially interesting to note in this study that all the post-treatment drawings were smaller than the 'before cancer' drawings. It could have been hypothesised that the post-treatment drawings would have been larger than the pre-cancer ones, allowing for greater area to depict areas of change. However, as this was not the case, it may be that the respondents were fatigued following the completion of the questionnaires and the first drawing and the second one was undertaken more quickly. The authors do not think this is the case, due to the detail added to the second picture. Further investigation with a larger sample may find a relationship with the severity of the tumour or the nature of the treatment regimen.

It has been suggested that drawings can uncover multiple dimensions of living with disease, especially psychosocial,³⁰ and this can provide healthcare professionals with a suggestion of how the patient is coping with their illness. This creativity involved in drawing offers patients a way to express themselves, which can minimise healthcare professionals' imposition of their own views. Drawing can also have potential benefits for patients. Drawing can be an informal opportunity to offer time and space for reflection; for some patients, this activity can access perceptions and emotions which may have been unknown previously. The uncovering of buried or unacknowledged aspects that may be causing distress could help patients better understand their post-treatment selves and needs. Not all patients will benefit from or be comfortable with one technique. However, drawing is another way for people, especially those who are visually or creatively orientated, to represent themselves.

Conclusion

This study suggests that drawing elicit distinctly different information and understanding about a person's body image following treatment for HNC than provided by HRQoL questionnaires. Further research is needed to clarify if clinical opinion matches the patients' drawing and if drawings would be feasible in a clinical appointment.

Acknowledgements

The authors would like to thank Dr T.P. Moss for his support with the development of this research.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

Ethical approval for this study was obtained from local (Bristol) NHS regulatory body (REC reference no. 10/H0107/24).

Funding

The author(s) disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: The project was self-funded by the lead author.

Informed consent

Written informed consent was obtained from all subjects before the study.

ORCID iD

Sam Harding  <https://orcid.org/0000-0002-5870-2094>

References

- Arora NK, Gustafson DH, Hawkins RP, et al. Impact of surgery and chemotherapy on the quality of life of younger women with breast carcinoma: a prospective study. *Cancer* 2013; 92: 1288–1298.
- Melissant HC, Neijenhuijs KI, Jansen F, et al. A systematic review of the measurement properties of the Body Image Scale (BIS) in cancer patients. *Support Care Cancer* 2018; 26(6): 1715–1726.
- Dropkin MJ. Body image and quality of life after head and neck cancer surgery. *Cancer Pract* 1999; 7(6): 309–313.
- Rogers SN, Fisher SE and Woolgar JA. A review of quality of life assessment in oral cancer. *Int J Oral Maxillofac Surg* 1999; 28: 99–117.
- Katre C, Johnson IA, Humphris GM, et al. Assessment of problems with appearance, following surgery for oral and oropharyngeal cancer using the University of Washington appearance domain and the Derriford appearance scale. *Oral Oncol* 2008; 44(10): 927–934.
- Clarke SA, Newell R, Thompson A, et al. Appearance concerns and psychosocial adjustment following head and neck cancer: a cross-sectional study and nine-month follow-up. *Psychol Health Med* 2014; 19(5): 505–518.
- Carr T, Moss T and Harris D. The DAS24: a short form of the Derriford Appearance Scale DAS59 to measure individual responses to living with problems of appearance. *Br J Health Psychol* 2005; 10(Pt 2): 285–298.
- Rogers SN, Gwanne S, Lowe D, et al. The addition of mood and anxiety domains to the University of Washington quality of life scale. *Head Neck* 2002; 24(6): 521–529.
- Weymuller EA Jr, Alsarraf R, Yueh B, et al. Analysis of the performance characteristics of the University of Washington Quality of Life instrument and its modification (UW-QOL-R). *Arch Otolaryngol Head Neck Surg* 2001; 127(5): 489–493.
- Scott SE, Birt L, Cavers D, et al. Patient drawings of their melanoma: a novel approach to understanding symptom perception and appraisal prior to healthcare. *Psychol Health* 2015; 30: 1035–1048.
- Edgar IR. The imagework method in health and social science research. *Qual Health Res* 1999; 9(2): 198–211.
- Kaptein AA and Broadbent E. Illness cognition assessment. In: Ayers S, Baum A, McManus C, et al. (eds) *Cambridge handbook of psychology, health and medicine*. 2nd ed. Cambridge: Cambridge University Press, 2007, pp. 268–273.
- Broadbent E, Ellis CJ, Gamble G, et al. Changes in patient drawings of the heart identify slow recovery after myocardial infarction. *Psychosom Med* 2006; 68(6): 910–913.
- Harrow A, Wells M, Humphris G, et al. 'Seeing is believing, and believing is seeing': an exploration of the meaning and impact of women's mental images of their breast cancer and their potential origins. *Patient Educ Couns* 2008; 73(2): 339–346.
- Bolwerk A, Mack-Andrick J, Lang FR, et al. How art changes your brain: differential effects of visual production and cognitive art evaluation on functional brain connectivity. *PLoS ONE* 2014; 9: e101035.
- Angstrom-Brannstrom C and Norberg A. Children undergoing cancer treatment describe their experiences of comfort in interviews and drawings. *J Pediatr Oncol Nurs* 2014; 31(3): 135–146.
- Cheung MMY, Saini B and Smith L. Using drawings to explore patients' perceptions of their illness: a scoping review. *J Multidiscip Healthc* 2016; 9: 631–646.
- Cancer Research UK. Oral cancer statistics. *Cancer Research UK*, <http://www.cancerresearchuk.org/health-professional/cancer-statistics/statistics-by-cancer-type/oral-cancer> (accessed 21 October 2018).
- Communities and Neighbourhoods. The English indices of deprivation 2007 (Rep. No. 07 NRAD 05137), <http://geoconvert.mimas.ac.uk/help/ind-2007-manual.pdf>
- Djan R and Penington A. A systematic review of questionnaires to measure the impact of appearance on quality of life for head and neck cancer patients. *J Plast Reconstr Aesthet Surg* 2013; 66(5): 647–659.
- Hassan SJ and Weymuller EA Jr. Assessment of quality of life in head and neck cancer patients. *Head Neck* 1993; 15: 485–496.
- National Institutes of Health. ImageJ, <http://rsbweb.nih.gov/ij/> (2009, accessed 25 April 2018).
- Broadbent E, Niederhoffer K, Hague T, et al. Headache sufferers' drawings reflect distress, disability and illness perceptions. *J Psychosom Res* 2009; 66(5): 465–470.
- Guillemin M. Understanding illness: using drawings as a research method. *Qual Health Res* 2004; 14(2): 272–289.
- Michie S and Abraham C. Interventions to change health behaviors: evidence-based or evidence inspired? *Psychol Health* 2004; 19: 29–49.
- Moore RJ and Spiegel D. Uses of guided imagery for pain control by African-American and White women with metastatic breast cancer. *Integr Med* 2000; 2(2): 115–126.
- Monti DA, Peterson C, Kunkel EJ, et al. A randomized, controlled trial of mindfulness-based art therapy (MBAT) for women with cancer. *Psychooncology* 2006; 15(5): 363–373.
- Collie K, Bottorff JL and Long BC. A narrative view of art therapy and art making by women with breast cancer. *J Health Psychol* 2006; 11(5): 761–775.
- Machover K. *Personality Projection in the drawing of the human figure: a method of personality investigation*. Springfield, IL: Charles C Thomas, 1971.
- Nowicka-Sauer K. Patients' perspective: lupus in patients' drawings. Assessing drawing as a diagnostic and therapeutic methods. *Clin Rheumatol* 2007; 26: 1523–1525.

Appendix 23: Heather van der Lely Foundation Trust Project Summary

Background

Developmental Language Disorder has a childhood prevalence rate of approximately 7%. Every UK primary classroom has children with difficulties understanding the complex instructions, formulating sentences, learning new vocabulary or making themselves understood. Developmental Language Disorder is associated with considerable risk to the child's future wellbeing, mental health and social integration.

Positive effects of early interventions have been demonstrated despite challenges in identification in preschool. Approaches are often known as Parent-Child Interaction Therapy (PCIT). The methods of delivery vary, however, the emphasis is on teaching the parents key strategies.

There are concerns about the validity of PCIT for some families. The inclusion of mostly westernised middle class families in studies underpinning PCIT limits their external validity. There are implicit cultural biases contained within PCIT, with assumptions about aspects of social organisation related to interaction, the value of talk, how status is handled in interaction, beliefs about intentionality, and beliefs about teaching language to children.

This lack of external validity could explain difficulties experienced by speech and language therapists trying to deliver PCIT to families from diverse backgrounds.

This programme of work aims to explore;

- the pattern of children's typical activities at home,
- what language interactions occur on a typical day,
- the relationship between beliefs and attitudes towards raising children and linguistic interactions with children

The objective is to improve understanding of typical interactions in preschool children from a range of backgrounds in order to facilitate the future development of theory of PCIT so that interventions are more appropriate and acceptable.

Method

Recruitment

Fifteen-to-twenty preschool children and their main caregivers, will be recruited purposively. Children will have a range of language development levels within the typical range, based on data collected via the preschool settings from the Early Years Foundation stage and the communication sections Ages & Stages Questionnaire. Children will be between 2 and 4 years 11 months.

Data collection:

Audio recordings will be captured during one 16 hour period for one typical day using the Language ENvironment Analysis (LENATM).

Diaries of family activity: Care-givers will be asked to keep a diary of the day that LENA is used.

Interviews with care-givers will be conducted to capture caregivers' immediate recollections of how these compare to other days; a second in-depth interview will explore caregivers' explanations of activities and structures of conversations.

Analysis:

Automated analysis of LENA recordings provides information on the interactions between parent and child that can be mapped onto activities. Times of high and low interaction will be analysed using discourse analysis. Non-English samples will be translated into English.

Synthesis:

The multiple data sets will be synthesised using a meta-ethnographic method; 'lines-of-argument'. This will build a rich description of communication environments that relates language patterns to social events and provides an understanding of the parental beliefs that underpin them.

Impact and dissemination:

Findings will recommend changes to existing PCIT and generate ideas for new ways of working that are sensitive to different contexts. This will enable development of guidelines for professional groups to support the delivery of PCIT interventions that are culturally sensitive to the range of families accessing health and education services.