




Please cite the Published Version

Mbada, Chidozie Emmanuel , Fashote, Emmanuel Odunayo, Awotidebe, Taofeek Oluwole, Olorunmoteni, Oluwatosin Eunice, Adebambo, Atitlola, Ademoyegun, Adekola B, Sonuga, Ademola O, Fatoye, Clara, Gebrye, Tadesse  and Fatoye, Francis  (2024) Development and Validation of a Tool to Assess Pain in Preverbal and Non-verbal Children with Cerebral Palsy: Cerebral Palsy Faces Pain Scale (CPFPS). *Rehabilitacja Medyczna*, 28 (1). pp. 4-12. ISSN 1427-9622

DOI: <https://doi.org/10.5604/01.3001.0054.7727>

Publisher: Index Copernicus

Version: Published Version

Downloaded from: <https://e-space.mmu.ac.uk/636638/>

Usage rights:  [Creative Commons: Attribution-Share Alike 4.0](https://creativecommons.org/licenses/by-sa/4.0/)

Additional Information: This is an open access article which first appeared in *Rehabilitacja Medyczna*

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>)

Development and Validation of a Tool to Assess Pain in Preverbal and Non-verbal Children with Cerebral Palsy: Cerebral Palsy Faces Pain Scale (CPFPS)

Chidozie Emmanuel Mbada¹ (A,B,C,D,E,F), Emmanuel Odunayo Fashote² (B,D,E,F),
Taofeek Oluwale Awotidebe³ (B,C,D,E,F), Oluwatosin Eunice Olorunmoteni⁴ (B,D,E,F),
Atitlola Adebambo⁵ (B,D,E,F), Adekola B. Ademoyegun^{3,6} (B,C,D,E,F), Ademola O. Sonuga³ (B,D,E,F),
Clara Fatoye⁷ (B,D,E,F), Tadesse Gebrye¹ (B,D,E,F), Francis Fatoye¹ (B,C,D,E,F)

¹ Department of Health Professions, Faculty of Health and Education, Manchester Metropolitan University, United Kingdom

² Children and Young People's Therapy, Hertfordshire Community Children Hospital, Peace Children Centre, Watford, United Kingdom

³ Department of Medical Rehabilitation, Obafemi Awolowo University, Ile-Ife, Nigeria

⁴ Department of Paediatrics and Child Health, Obafemi Awolowo University, Ile-Ife, Nigeria

⁵ Children's Physiotherapy Department, Kaleidoscope Centre, Rushey Green, Catford London, United Kingdom

⁶ Department of Physiotherapy, Osun State University Teaching Hospital, Osogbo, Nigeria

⁷ Faculty of Health and Care Professions, University Campus Oldham, Oldham, United Kingdom

Keywords

developmental milestones, paediatrics, facial expression, psychometrics

Abstract

Background: Cerebral palsy (CP) is the most common neurodisability in children. This study aimed to develop and validate a faces pain scale for preverbal and non-verbal children (PvNv) with CP in Nigeria.

Methods: Twelve paediatric practitioners and three verbal adults with CP participated in a three-round modified Delphi study on scale development and validation of the CPFPS. Data from the scale development were analysed using the items content validity index (I-CVI) and scale content validity (S-CVI). The concurrent validity of CPFPS was established using the University of Wisconsin Children's Hospital Pain Scale (UWHPS) for PvNv children.

Results: Facial characteristics such as furrowing, crying, clenching and grinding of teeth, quivering lips, and changes in the eye may indicate pain in PvNv CP. These features have I-CVI scores of 1.00, 1.00, 0.83, 0.83, and 0.83, respectively. Moreover, the "eye/furrow" and "mouth/nasolabial" features were identified as sub-scales with S-CVI scores of 1.00 and 0.83, respectively, while the CPFPS received an S-CVI score of 0.89. A significant positive correlation was observed between CPFPS and the UWHPS ($\rho=0.949$; $p=0.001$) (concurrent validity). CPFPS also exhibited high inter-rater reliability (ICC=0.994, 95% CI=0.988-0.997). However, the known-group validity of the CPFPS with age, CP types and functional status were non-significant ($p>0.05$), except for the communication ability of children with CP ($\rho=0.233$; $p=0.022$).

Conclusions: CPFPS, a pain assessment tool using "eye/furrow" and "mouth/nasolabial" features, is reliable for evaluating pain in PvNv children with CP in Nigeria. Our results confirm the initial validity and reliability of the CPFPS, but further investigations into the evaluation of other psychometric properties are needed.

INTRODUCTION

Living with cerebral palsy (CP) is associated with significant challenges in cognition and development which, in turn, impact an individual's quality of life^{1,2}. Despite the physical limitations in movement

and posture, as well as intellectual and developmental disabilities, children with CP experience more pain compared to their typically-developing peers^{1,3}. The higher rates and intensity of pain in children with CP are due to their chronic physical impairment, associated medical condi-

tions, increased frequency of injuries and the toll of medical interventions³⁻⁵. In previous studies, it has been estimated that 83% of children with CP experience pain, with nearly 50% of them affected by some form of pain either all the time or at some point of each day^{5,6}.

The individual division of this paper was as follows: A – research work project; B – data collection; C – statistical analysis; D – data interpretation; E – manuscript compilation; F – publication search

Article received: 27 Jan. 2024 / accepted: 25 Sep. 2024

Please cite as: Mbada Ch.E., Fashote E.O., Awotidebe T.O., Olorunmoteni O.E., Adebambo A., Ademoyegun A.B., Sonuga A.O., Fatoye C., Gebrye T., Fatoye F. Development and Validation of a Tool to Assess Pain in Preverbal and Non-verbal Children with Cerebral Palsy: Cerebral Palsy Faces Pain Scale (CPFPS). Med Rehabil 2024; 28(2): 4-12. DOI: 10.5604/01.3001.0054.7727

Internet version (original): www.rehmed.pl

This article is licensed under the Creative Commons Attribution-ShareAlike 4.0 International License CC BY-SA (<http://creativecommons.org/licenses/by-sa/4.0/>)

The assessment of pain experience in children with CP poses a significant clinical challenge, particularly, as many of them may have difficulty expressing themselves due to their motor impairment². Other medical complications such as visual impairments⁷ and mental retardation, which affects about 40% of children with CP, can also have negative impact on their intelligence quotient⁸. Despite the cognitive challenges involved in assessing pain among children with CP, in previous studies, proxies such as clinicians^{9,10} and parental reports have been used^{11,12}. While clinicians' assessment of pain is considered more suitable for acute pain, parental or primary caregiver proxies are preferred for chronic pain assessment¹¹. However, there is often a significant difference between pain scores reported by clinicians and caregivers^{13,14}. Additionally, parental proxies have limited use in institutionalised children with CP¹⁵. While clinicians' and caregivers' reports are still used as proxies, they are only useful for a small population of children with CP due to maladaptive behaviours³. Observational measures may also offer guidance for assessing pain in non-verbal populations with CP³.

Following a cursory search of the available literature on pain assessment in CP, it is apparent that there is a paucity of pain assessment tools tailored to non-verbal children with CP. Specifically, there is not existing scale to evaluate pain in African children with non-verbal CP. Therefore, the need arises to develop a culturally sensitive and specific pain assessment tool for non-verbal children with CP. Based on recommendations from previous studies indicating the use of pain faces to deduce pain intensity in non-verbal populations¹⁶, a study to develop a new pain tool incorporating the faces of African children with CP was conducted. In the design of the pain tool, this study tracked similar studies such as the Faces Pain Scale¹⁷, the University of Wisconsin Children's Hospital Pain Scale (UWHPS) for Preverbal and Nonverbal (PvNv) children¹⁸, the Pain Assessment Instrument for Cerebral Palsy (PAICP)¹⁹ and the Caregiver

Priorities and Child Health Index of Life with Disabilities Questionnaire¹². Features for depicting pain common to those in the studies given above include non-verbal vocalisations, facial grimacing, bracing, rubbing, restlessness and vocal complaints. In addition to exploring these features in the development of the new tool, an important consideration based on anecdotes and literature is that the African culture inhibits pain expression, and there is less facial expression of pain experienced compared to children from other contexts²⁰. This study aimed to develop and validate a cultural-friendly faces pain scale for PvNv with CP in Nigeria.

MATERIALS AND METHODS

In this study, a mixed methods model was used to develop and validate the Cerebral Palsy Faces Pain Scale (CPFPS).

Development phase of the study

In this phase, a modified Delphi approach was used to identify content for assessing pain in children with CP²¹. In Delphi studies, a minimum sample of 12 participants is usually considered sufficient to achieve consensus, whereas having larger sample sizes results in diminishing returns regarding validity of the findings²². Accordingly, in this study, 12 healthcare practitioners from the Obafemi Awolowo University Teaching Hospital Complex (OAUTHC) in Ile-Ife, Osun State, Nigeria (three nurses, Occupational Therapists, Physicians and Physiotherapists, respectively) were recruited. These healthcare professionals were experts in CP care. To be eligible for the study, respondents had to meet one of the following criteria: be a fellow of the Postgraduate Medical College with a specialisation in paediatrics, hold a master's or doctorate in a field related to paediatrics, have evidence of publications on CP, pain assessment, or scale development, and possessing work experience of more than five years in CP management. In this

study, nine out of the 12 Delphi participants were male, while nine and three participants have had 5-9 years and > 10 years of experience in paediatric practice. In all, a consultant paediatrician, two residents in paediatrics, one assistant director of physiotherapy, four principals (one nurse, one physiotherapist, and two occupational therapists), and four chiefs (two nurses, one physiotherapist, and one occupational therapist) were involved.

Additionally, three verbal adults with CP, who were 18 years of age or older and with no other co-morbidities, were also part of the study.

The modified Delphi technique aims to gather consensus from experts with the highest item content validity index (I-CVI). The I-CVI of an item is calculated by dividing the number of reviewers giving a rating of three or four ('moderately relevant' or 'very relevant') for that item by the total number of reviewers²³. In this study, the modified Delphi method involved three rounds. In the first round, an open-ended questionnaire was distributed to 12 healthcare practitioners, asking them to identify clinical factors or features that can effectively indicate the presence of pain in children with CP. Data from round one were summarised and presented based on evaluation forms completed by the 12 reviewers. In the second round, the experts were asked to rate the proposed items' relevance using a 4-point Likert scale (1 = 'not relevant', 2 = 'relevant, needs major revision', 3 = 'moderately relevant, needs minor revision', and 4 = 'very relevant, no modification'). They were also asked to identify which photographs (from several showing pain faces of children with CP) could be used for a scale aimed at depicting pain. In the third stage of the study, the items were sent to the experts with their respective I-CVI ratings. Suggestions from the second stage were also sent to the experts for deliberation and possible consensus. Feedback from three adults with CP was sought to provide user input to the final version of the pilot tool. To ascertain the face validity of the new face scale, three verbal adults

living with CP, who were not part of the scale development process, were asked to ascribe the severity of pain to the face photographs.

Validation phase of the study

The objective of this phases was to assess the concurrent validity, known-group validity and inter-rater reliability of the CPFPS. During the validation phase of the study, we recruited children with CP aged between six months and 10 years. The participants were non-verbal patients with CP who were attending outpatient appointments, and the clinicians involved included had experience in paediatric care for children with CP. The main inclusion criteria for the patient group were a physician diagnosis of CP and a report of absence of meaningful words, in addition to the use of the Communication Function Classification System (CFCS). The CFCS is a tool used to classify children with CP based on their communicative abilities and has been documented in the literature to demonstrate good psychometric properties²⁴. The CFCS was employed in this study to describe and classify the communication ability of the sample. The functional status of the patients was also assessed using the Gross Motor Function Classification System (GMFCS) and the Communication Function Classification System (CFCS). The GMFCS was developed in accordance with the International Classification of Functioning, Disability and Health (ICF) philosophy to record the functional abilities and limitations that children with CP experience in their daily lives²⁵. The type of CP was also assessed based on the International Statistical Classification of Diseases and Related Health Problems 10th Revision (ICD-10).

The concurrent validity of the pain tool (CPFPS) was established using the UWHPS for Pvnv children¹⁸. The UWHPS for Pvnv children was developed and tested on preverbal children (below the age of three) and cognitively impaired children to assess pain but not that specific to CP. The psychometric properties of the scale were adjudged acceptable. The inter-

nal consistency of the scale (Cronbach's alpha) and inter-rater reliability were 0.93 and 0.92, respectively. The correlation of the UWHPS for Pvnv children with the Wong-Baker Faces Scale was $r=0.62$ ¹⁸. Inter-observer reliability of the new scale among clinicians and parents of children with Pvnv CP was tested by two raters who were familiar with the use of the different scales (Assessors: Emmanuel Fashote and Atilola Adebambo) evaluating the reproducibility of the scale. Spearman's rank correlation coefficient was used to investigate the known-group validity of the CPFPS by correlating the tool with age and CP parameters.

According to de Vet et al., a sample size of 50 individuals is required, anticipating an ICC of 0.8 and a confidence interval of 95% ($95\% CI \pm 0.1[0.7-0.9]$)²⁶. Also, the Consensus-based Standards for the selection of health status Measurement Instruments (COSMIN) recommend the sample size be at least 100 for validity testing of patients' reported outcome measures²⁷. In accordance, a sample size of 75 participants for both clinicians and parents was adjudged as sufficient to give the lowest expected correlation coefficient of $r=0.50$ with the Alpha of $p<0.05$ and power of 80%²⁸. Ethical approval for this study was obtained from the Ethics and Research Committee of the OAUTHC, Ile-Ife, Nigeria (ERC/2020/03/02).

Data analysis

The Delphi phase data was analysed using the I-CVI method to identify themes. An item's I-CVI is calculated by dividing the number of reviewers who rated the item as 'moderately relevant' or 'very relevant' (three or four) by the total number of reviewers²³. A minimum I-CVI of ≥ 0.78 was considered valid based on the number of experts on the review panel²⁹. Descriptive statistics, including means, standard deviations and percentages, were used to summarise the data. The CPFPS implemented metric scoring of 0 to 10, and closely follows a linear interval scale, thus, Spearman's rank correlation coefficient and In-

tra-Class Correlation (ICC) were used to assess its concurrent validity and inter-rater reliability. The ranges of the correlation were scores considered as follows: poor <0.5 ; moderate 0.5-0.75; good 0.75-0.9 and excellent >0.9 ³⁰. Spearman's rank correlation coefficient was also applied to test the known-group validation of the CPFPS. Known-group validity is the instrument's ability to differentiate among distinct groups³¹. A scatter plot was employed to illustrate the validity of the CPFPS. Data was analysed using the Statistical Program for Social Sciences for Windows version 22 (SPSS Inc., Chicago, Illinois, United States). The alpha level was set at $p<0.05$.

RESULTS

Development phase of the study

A CP faces pain scale (CPFPS) for CP was developed. CPFPS utilises "eye/furrow" and "mouth/nasolabial" features that may indicate pain in Pvnv CP (Figure 1). The 16 items suggested by the Delphi panel after the first round of the Delphi with their I-CVI values are shown in Table 1. The items generated in the first round were grouped under four themes: vital signs, facial, behaviour and body movement (Table 2). There was a 90% consensus on the photographs of the pain faces sent to the experts for the face part of the tool. The experts also suggested that the items with I-CVI > 0.78 are the subset of the facial theme in round one of the Delphi and should, therefore, be included in the pain faces scale. Three experts suggested that the items be scaled in a simple metric of 0-10. One of the experts, a nurse, recommended the pain face be divided into two scales: the eye furrow scale and the mouth-nasolabial scale. Items with I-CVI >0.78 and the distribution of the items into eye-furrow scale and mouth-nasolabial scale, correspondingly, are demonstrated in Table 1. There was a 100% consensus on having the tool in two scales, as suggested by one of the experts. Some of the quotes used by the experts are given in Table 2.

NAME: _____

AGE: _____













CEREBRAL PALSY FACES PAIN SCALE		
	EYE/FURROW SCALE	MOUTH/NASOLABIAL SCALE
0 No pain at all		
2		
4		
6		
8		
10 Most severe pain		

Figure 1
Cerebral Palsy Faces Pain Scale

Table 1

Items' suggestions from Round 1 and the respective I-CVI		
S/N	Items pool	I-CVI
1	Temperature	0.08
2	Respiratory Rate	0.08
3	Pulse Rate	0.17
4	Furrow* ^E	1.00
5	Crying* ^E	1.00
6	Clenching and grinding of teeth* ^M	0.83
7	Lips quivering* ^M	0.83
8	Changes in Eyes* ^E	0.83
9	Panics	0.33
10	Poor sucking	0.33
11	Inconsolable	0.33
12	Poor Cooperation	0.42
13	Withdrawal	0.42
14	Muscle Tone	0.33
15	Postural Imbalance	0.17
16	Voluntary Immobility	0.25

I-CVI – Item Content Validity Index; S-CVI – Scale Content Validity Index; * – I-CVI score > 0.78; ^E – Items categorised as eye-furrow scale from round 2; ^M – Items categorised as mouth-nasolabial from round 2; S-CVI^E - 1.00+1.00+0.83/3=0.94; S-CVI^M - 0.83+0.83/2=0.83

Table 2

Themes from the first round of Delphi phase of the study with selected quotes			
Theme	Items	Experts	Quotes
Vital Signs	Temperature	Expert 9 (physician)	"Respiratory rate often changes with pain"
	Respiratory rate		
	Pulse rate		
Facial	Furrow	Expert 2 (physiotherapist)	"furrow always point to point"
	Crying		
	Clenching and grinding of teeth		
	Lips quivering		
Behavior	Changes in eye	Expert 5 (Nurse)	"when they turn down the breast, there is pain"
	Poor sucking		
	Inconsolable		
	Poor cooperation		
Body movement	Withdrawal	Expert 11 (Occupational therapist)	"pain is accompanied by abnormal tone"
	Muscle tone		
	Postural imbalance		
	Voluntary immobility		
Quotes showing opinion of experts on having the tool as two scales			
Experts		Quotes	
Expert 2 (Physician)		"this would make the tool more detailed in assessing the facial features of pain expression"	
Expert 7 (Physiotherapist)		"scoring pain under both scales would cater for differences in facial expression"	
Expert 10 (Nurse)		"we may not miss out any signal of pain with this idea"	
Expert 12 (Occupational therapist)		"this would make the tool different from conventional face pain tools"	

CPFPS has two subscales based on "eye/furrow" and "mouth/nasolabial" features. Each subscale uses a metric scoring from 0 ('no pain at all') to 10 ('most severe pain'), and either can be used to describe pain. The chosen faces based on "eye/furrow" and "mouth/nasolabial" features are scored 0, 2, 4, 6, 8, or 10. The faces show increasing pain from top to bottom and correspond with the scores. "Eye/furrow" subscale features of pain are presented on the left, while the "mouth/nasolabial" features are presented on the right. The child would then choose the image that corresponds to the level of pain they experience, and the corresponding score is recorded.

Validation phase of the study

Socio-demographic characteristics such as age and sex distributions of the participants in the validation phase of the study are shown in Table 3. The mean age of the participants was 17.55 ± 9.75 months, ranging be-

tween six and 97 months. Spastic diplegia was found to be the most prevalent type of CP with an outcome of 36 (36.0%) and spastic triplegia being the least, in two cases (2.0%). The distribution of the participants' CP parameters is presented in Table 3. For concurrent validity, a positive inter-rater reliability was observed between CPFPS and the UWHPS ($\rho=0.949$; $p=0.001$). CPFPS also exhibited high inter-rater reliability (ICC=0.994, 95% CI=0.988-0.997) (Figure 2, Table 4). The results indicate no significant correlations between CPFPS and age ($\rho=0.026$, $p=0.801$), GMFCS ($\rho=0.099$, $p=0.338$), or CP types ($\rho=-0.050$, $p=0.628$) (Table 5), except for CFCS ($\rho=0.233$, $p=0.022$).

DISCUSSION

The study aimed to develop a CPFPS for PvNv with CP, determine the concurrent validity of the CPFPS using the UWHPS, and examine the inter-rater reliability of the tool. Follow-

ing a modified Delphi approach, the CPFPS was developed. The tool has two subscales based on "eye/furrow" and "mouth/nasolabial" features. Each subscale uses a metric scoring from 0 ('no pain at all') to 10 ('most severe pain'), and either can be used to depict pain in PvNv children with CP. Faces reflect how much something can hurt³². As a result, face pain scales are conventionally used by healthcare providers as one of the methods for pain assessment in children by employing scales of different facial expressions, and linking their experience to a related face³². A number of faces pain scales have been developed with moderate to high psychometric properties. These include UWHPS¹⁸, The Sydney Animated Facial Expressions (SAFE)³³, Faces Pain Scale¹⁷, Faces Pain Scale-Revised (FPS-R)³², the African-American Oucher pain scale³⁴ and Wong-Baker Faces Pain Rating Scale (WBFPR)³⁵. Some of these scales are generic in nature, and can be adapted to different conditions, while others are disease-specific³⁶. Also, face

Table 3

Socio-demographic and clinical characteristics with distribution of cerebral palsy parameters of the participants		
Variable	Frequency	Percentage
Sex		
Male	50	50.0
Female	50	50.0
Age group (months)		
6–15	56	56.0
16–25	31	31.0
26–35	3	3.0
36–45	8	8.0
Type		
Spastic diplegia	36	36.0
Spastic quadriplegia	25	25.0
Spastic hemiplegia	18	18.0
Spastic athetoid	16	16.0
Ataxic	3	3.0
Spastic triplegia	2	2.0
GMFCS Rating		
1	3	3.0
2	8	8.0
3	24	24.0
4	45	45.0
5	20	20.0
CFCS Rating		
2	3	3.0
3	27	27.0
4	35	35.0
5	35	35.0

GMFCS – Gross Motor Function Classification System; CFCS – Communication Function Classification System

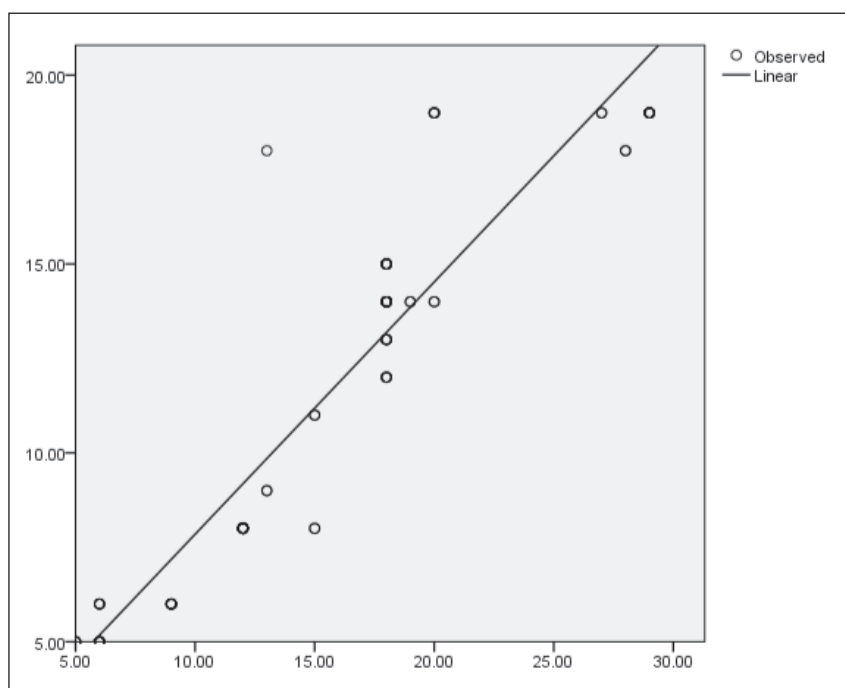


Figure 2
A scatter plot graph showing the correlation between the scores on the Cerebral Palsy Faces Pain Scale and the University of Wisconsin Hospital Pain Scale

scales are conventionally preferred, especially by younger children³⁷. It is believed that children are better able to rate the level of pain they experience using face ratings compared to verbal or numerical ratings^{38,39}. Observations also note that children who are five and older can provide reliable reports of the pain they experience³⁶. It is also reported that faces pain scales are generally appealing to children and are fairly quick as well as easy to administer⁴⁰. Tomlinson et al.³⁶ in a systematic review of faces scales for the self-report of pain intensity in children, posits that it remains unclear whether any one of the faces scales is better for a particular purpose based on psychometric properties and preferences.

Assessing pain experienced by children with CP is particularly difficult owing to complications arising from motor, visual and verbal impairments¹⁹. Thus, self-reported faces pain scales may have limited application among children with CP who cannot express their pain verbally⁴¹. While a number of CP-specific pain scales, such as Pain Assessment Instrument for Cerebral Palsy [PIACP]¹⁹, revised Face, Legs, Activity, Cry and Consolability (r-FLACC)⁴², and Individualised Numeric Rating Scale (INRS)⁴³ have been developed. However, the applicability of these scales among children with CP with severe disability or verbal impairment is limited. As a result, the common practice in situations involving these types of children is still to rely on caregiver interviews to assess the amount of pain experienced by the child^{44,45}. Unfortunately, in a number of studies, disparities have been reported between caregiver pain reports and pain experienced by children^{9,10,13,14,46,47}. Unfortunately, the pain scored by the patient often differs from the pain scored by the caregivers^{14,32,48}. Hence, a need for non-communicating children’s pain scales that are specific to CP.

CPFPS was designed as a scale for caregivers and healthcare providers to assess pain in PvNv children with CP. CPFPS adopts common metric practice in accordance with proposal by von Baeyer⁴⁹. Measures based on a common metric sys-

Table 4

Concurrent validity and inter-rater reliability test of the Cerebral Palsy Faces Pain Scale			
Variable	UWHPS		
Validity	P	p-value	
CPFPS	0.949	0.0001	
Reliability	95% CI	p-value	
	Lower - Upper		
Inter-rater reliability			
Patients – Parents	0.994	0.988 - 0.997	0.001

CPFPS – Cerebral Palsy Faces Pain Scale; UWHPS – University of Wisconsin Hospital Pain Scale; ρ – Spearman rank correlation coefficient; 95% CI – 95 percent confidence interval

Table 5

Known group validation of the Cerebral Palsy Faces Pain Scale								
Variable	Age		CP Type		GMFCS		CFCS	
	ρ	p-value	ρ	p-value	P	p-value	ρ	p-value
CPFPS	0.026	0.801	-0.05	0.628	0.099	0.338	0.233	0.022*

* – Indicates significant correlation; CP – Cerebral Palsy Type; CFCS – Communication Function Classification System; CPFPS – Cerebral Palsy Faces Pain Scale; GMFCS – Gross Motor Function Classification System; ρ – Spearman rank correlation coefficient

tem have been reported as time efficient, simple to use, and enhance consistency between two users³². CPFPS adopts typical faces of sub-Saharan African children with CP, rather than using emoticons. Ethno-cultural differences have been reported to be a limitation of emoticon-based pain tools⁵⁰. As a result, researchers have recommended facial emoticons that are culturally specific⁵¹⁻⁵³. CPFPS was designed to depict pain experienced by PvNV children with CP based on “eye/furrow” and “mouth/nasolabial” features. Similarly, in previous tools such as the UWHPS, facial features involving forehead furrows and frown lines, as well as crying facial expressions have been used for depicting pain intensity in children with neurological deficits¹⁸. CPFPS has simplified these descriptors into pictures to make pain assessment fairly easier.

The CPFPS demonstrated excellent concurrent validity among 100 pa-

tients with CP. The concurrent validity was considered adequate ($\rho=0.949$; $p=0.001$), having scored a correlation coefficient >0.9 . UWHPS was used as the comparator for the concurrent validity because it is a scale designed to assess pain among PvNV children with neurological deficits¹⁸. CPFPS also shows excellent reliability when applied to depict the pain experiences of PvNV children with CP. According to the National Institute for Health and Care Excellence (NICE) guidelines⁵⁴, the validity, reliability and accuracy of pain measurement scales employed in the assessment of pain in CP are important considering the learning or communication impairments of the patients. Thus, it is implied that CPFPS can be used to assess pain in PvNV children with CP. The findings of this study are consistent with earlier reports in which it was noted that face scales are valid and reliable measures of pain intensity in children with CP^{35,55}.

The findings on known-group validity of the CPFPS with age, CP types and functional status indicate no significant correlations, except the communication ability of children with CP. This implies that CPFPS, as a pain tool, may not be able to discriminate between groups of PvNV children with CP known to differ on account of age, CP types and functional status. The significant correlation between CPFPS and CFCS may imply that the severity of communication impairment may influence pain assessment scores using the tool.

A potential limitation of this study is that the sample was drawn from only one public-funded tertiary hospital in Nigeria (OAUTHC, Osun State). However, the hospital is a referral centre for neighbouring Edo, Ekiti, Kwara, Lagos, and Ondo States. The sample in this study was diverse regarding types of CP, functional status and communication ability, which may enhance the scale’s applicability to a broader popu-

lation of PvNv children with CP. The CPFPS is designed and preliminarily validated to assess pain experienced by PvNv children with CP in Nigeria and in similar contexts. However, there is a need for validation of the CPFPS in similar and broader contexts among PvNv children with CP in line with the recommendations of the Consensus-based Standards for the selection of health Measurement Instruments (COSMIN)⁵⁶.

CONCLUSION

CPFPS, a pain assessment tool using “eye/furrow” and “mouth/nasolabial” features, is a reliable method for evaluating pain in PvNv children with CP in Nigeria. The CPFPS is strongly correlated with UWHPS, a validated pain descriptor scale for PvNv children. Our results confirm the initial validity and reliability of the CPFPS, but further investigations into the evaluation of other psychometric properties are needed.

List of abbreviations

CP – Cerebral palsy; PvNv – Preverbal and non-verbal; I-CVI – Items content validity index; S-CVI – Scale content validity index; CFCS – Communication Function Classification System; GMFCS – Gross Motor Function Classification System; UWHPS – University of Wisconsin Hospital Pain Scale; PA-ICP – Pain Assessment Instrument for Cerebral Palsy; CPFPS – Cerebral Palsy Faces Pain Scale; OAUTHC – Obafemi Awolowo University Teaching Hospital Complex; ICF – International Classification of Functioning, Disability and Health; ICD-10 – International Statistical Classification of Diseases and Related Health Problems 10th Revision; COSMIN – Consensus-based Standards for the Selection of Health Status Measurement Instruments; ICC – Intra-Class Correlation; CI – Confidence Interval; SAFE Sydney Animated Facial Expressions

FPS-R Faces Pain Scale-Revised; WBF-PR – Wong-Baker Faces Pain Rating Scale; r-FLACC – revised Face, Legs, Activity, Cry and Consol ability; INRS – Individualized Numeric Rating Scale; NICE – National Institute for Health and Care Excellence.

Approval of the Research Ethics Committee

Ethical approval for this study was obtained from the Ethics and Research Committee of the OAUTHC, Ile-Ife, Nigeria (ERC/2020/03/02).

Conflict of interest

The authors declare no conflict of interest.

References

- Novak I., Hines M., Goldsmith S., Barclay R. Clinical prognostic messages from a systematic review on cerebral palsy. *Pediatrics* 2012; 130(5): e1285-312. doi: 10.1542/peds.2012-0924.
- Olawale O.A., Deih A.N., Yaadar R.K. Psychological impact of cerebral palsy on families: The African perspective. *J Neurosci Rural Pract* 2013; 4(2): 159-63. doi: 10.4103/0976-3147.112752.
- Breau L.M., Burkitt C. Assessing pain in children with intellectual disabilities. *Pain Res Manag* 2009; 14(2): 116-20. doi: 10.1155/2009/642352.
- Breau L.M., Camfield C.S., McGrath P.J., Finley G.A. The incidence of pain in children with severe cognitive impairments. *Arch Pediatr Adolesc Med* 2003; 157(12): 1219-26. doi: 10.1001/archpedi.157.12.1219.
- Houlihan C.M., O'Donnell M., Conaway M., Stevenson R.D. Bodily pain and health-related quality of life in children with cerebral palsy. *Dev Med Child Neurol* 2004; 46(5): 305-10. doi: 10.1017/s0012162204000507.
- Tervo R.C., Symons F., Stout J., Novacheck T. Parental report of pain and associated limitations in ambulatory children with cerebral palsy. *Arch Phys Med Rehabil* 2006; 87(7): 928-34. doi: 10.1016/j.apmr.2006.02.023.
- Ipata A.E., Cioni G., Boldrini A. et al. Visual acuity of low- and high-risk neonates and acuity development during the first year. *Behav Brain Res* 1992; 49(1):107-14. doi: 10.1016/s0166-4328(05)80200-1.
- Nelson K.B., Grether J.K. Causes of cerebral palsy. *Curr Opin Pediatr* 1999; 11(6):487-91. doi: 10.1097/00008480-199912000-00002.
- Breau L.M., McGrath P.J., Camfield C. et al. Preliminary validation of an observational pain checklist for persons with cognitive impairments and inability to communicate verbally. *Dev Med Child Neurol* 2000; 42(9): 609-16. doi: 10.1017/s0012162200001146.
- Chibnall J.T., Tait J.C. Pain assessment in cognitively impaired and unimpaired older adults: a comparison of four scales. *Pain* 2001; 92(1-2): 173-86. doi: 10.1016/s0304-3959(00)00485-1.
- Hunt A., Mastroiannopoulou K., Goldman A., Seers K. Not knowing--the problem of pain in children with severe neurological impairment. *Int J Nurs Stud* 2003; 40(2): 171-83. doi: 10.1016/s0020-7489(02)00058-5.
- Jayanath S., Ong L.C., Marret M.J., Fauzi A.A. Parent-reported pain in non-verbal children and adolescents with cerebral palsy. *Dev Med Child Neurol* 2016; 58(4): 395-401. doi: 10.1111/dmcn.12943.
- Blomqvist K., Hallberg I.R. Pain in older adults living in sheltered accommodation--agreement between assessments by older adults and staff. *J Clin Nurs* 1999; 8(2): 159-69. doi: 10.1046/j.1365-2702.1999.00241.x.
- Garber J., Van Slyke D.A., Walker L.S. Concordance between mothers' and children's reports of somatic and emotional symptoms in patients with recurrent abdominal pain or emotional disorders. *J Abnorm Child Psychol* 1998; 26(5): 381-91. doi: 10.1023/a:1021955907190.
- Morrow A.M., Quine S., Loughlin E.V., Craig J.C. Different priorities: a comparison of parents' and health professionals' perceptions of quality of life in quadriplegic cerebral palsy. *Arch Dis Child* 2008; 93(2): 119-25. doi: 10.1136/adc.2006.115055.
- Chambers C.T., Reid G.J., Craig K.D. et al. Agreement between child and parent reports of pain. *Clin J Pain* 1998; 14(4): 336-42. doi: 10.1097/00002508-199812000-00011.
- Bieri D., Reeve R.A., Champion D.G. et al. The Faces Pain Scale for the self-assessment of the severity of pain experienced by children: development, initial validation, and preliminary investigation for ratio scale properties. *Pain* 1990; 41(2): 139-150. doi: 10.1016/0304-3959(90)90018-9.
- Soetenga D., Frank J., Pellino T.A. Assessment of the validity and reliability of the University of Wisconsin Children's Hospital Pain scale for Preverbal and Nonverbal Children. *Pediatr Nurs* 1999; 25(6): 670-6.
- Boldingh E.J., Jacobs-van der Bruggen M.A., Lankhorst G.J., Bouter L.M. Assessing pain in patients with severe cerebral palsy: development, reliability, and validity of a pain assessment instrument for cerebral palsy. *Arch Phys Med Rehabil* 2004; 85(5): 758-66. doi: 10.1016/j.apmr.2003.06.029.
- Nortje N., Albertyn R. The cultural language of pain: A South African study. *S Afr Fam Pract* 2015; 57(1): 24-27.
- Nasa P., Jain R., Juneja D. Delphi methodology in healthcare research: How to decide its appropriateness. *World J Methodol* 2021; 11(4): 116-129. doi: 10.5662/wjm.v11.i4.116.
- Vogel C., Zwolinsky S., Griffiths C. et al. A Delphi study to build consensus on the definition and use of big data in obesity research. *Int J Obes (Lond)* 2019; 43(12): 2573-2586. doi: 10.1038/s41366-018-0313-9.
- Polit D.F., Beck C.T. The content validity index: are you sure you know what's being reported? Critique and recommendations. *Res Nurs Health* 2006; 29(5): 489-97. doi: 10.1002/nur.20147.
- Hidecker M.J., Paneth N., Rosenbaum P.L. et al. Developing and validating the Communication Function Classification System for individuals with cerebral palsy. *Dev Med Child Neurol* 2011; 53(8): 704-10. doi: 10.1111/j.1469-8749.2011.03996.x.
- Palisano R.J., Rosenbaum P., Bartlett D., Livingston M.H. Content validity of the expanded and revised Gross Motor Function Classification System. *Dev Med Child Neurol* 2008; 50(10): 744-50. doi: 10.1111/j.1469-8749.2008.03089.x.
- de Vet H.C.W., Terwee C.B., Mokkink L.B., Knol D.L. *Measurement in Medicine*. 1st ed. Cambridge: Cambridge University Press; 2011.
- Terwee C.B., Prinsen C.A.C., Chiarotto A. et al. (2018). COSMIN methodology for assessing the content validity of PROMs. User manual version 1.0. 2018. www.cosmin.nl.
- Bland J.M., Altman D.G. Statistical methods for assessing agreement between two methods of clinical measurement. *Lancet* 1986; 1(8476): 307-10.
- Lynn M.R. Determination and quantification of content validity. *Nurs Res* 1986; 35(6): 382-5.
- Portney L.G., Watkins M. P. *Foundations of Clinical Research Applications to Practice*. Pearson/Prentice Hall, 2009.
- McConnell S., Kolopack P., Davis A.M. The Western Ontario and McMaster Universities Osteoarthritis Index (WOMAC): a review of its utility and measurement properties. *Arthritis Rheum* 2001; 45(5): 453-61. doi: 10.1002/1529-0131(200110)45:5<453::aid-art365>3.0.co;2-w.
- Hicks C.L., von Baeyer C.L., Spafford P.A. et al. The Faces Pain Scale-Revised: toward a common metric in pediatric pain measurement. *Pain* 2001; 93(2): 173-183. doi: 10.1016/S0304-3959(01)00314-1.
- Goodenough B., Thomas W., Champion G.D., Perret et al. Unravelling age effects and sex differences in needle pain: ratings of sensory intensity and unpleasantness of venipuncture pain by children and their parents. *Pain* 1999; 80(1-2): 179-90. doi: 10.1016/s0304-3959(98)00201-2.
- Beyer J.E., Aradine C.R. Content validity of an instrument to measure young children's perceptions of the intensity of their pain. *J Pediatr Nurs* 1986; 1(6): 386-95.
- Wong D.L., Baker C.M. Pain in children: comparison of assessment scales. *Pediatr Nurs* 1988; 14(1): 9-17.
- Tomlinson D., von Baeyer C.L., Stinson J.N., Sung L. A systematic review of faces scales for the self-report of pain intensity in children. *Pediatrics* 2010; 126(5): e1168-98. doi: 10.1542/peds.2010-1609.

37. de Tovar C., von Baeyer C.L., Wood C. et al. Postoperative self-report of pain in children: interscale agreement, response to analgesic, and preference for a faces scale and a visual analogue scale. *Pain Res Manag* 2010; 15(3): 163-8. doi: 10.1155/2010/475907.
38. Champion G.D., Goodenough B., Chua K. et al. Children's self-report of pain intensity using six different scale types. Paper presented at the 6th International Symposium on Pediatric Pain, June 2000, London.
39. Miró J., Castarlenas E., Huguet A. Evidence for the use of a numerical rating scale to assess the intensity of pediatric pain. *Eur J Pain* 2009; 13(10): 1089-95. doi: 10.1016/j.ejpain.2009.07.002.
40. Hunter M., McDowell L., Hennessy R., Cassey J. An evaluation of the Faces Pain Scale with young children. *J Pain Symptom Manage* 2000; 20(2): 122-9. doi: 10.1016/s0885-3924(00)00171-8.
41. Srivastava V.K., Laisram N., Srivastava R.K. Cerebral palsy. *Indian Pediatr* 1992; 29(8): 993-6.
42. Pedersen L.K., Rahbek O., Nikolajsen L., Møller-Madsen B. The revised FLACC score: Reliability and validation for pain assessment in children with cerebral palsy. *Scand J Pain* 2015; 9(1): 57-61. doi: 10.1016/j.sjpa.2015.06.007.
43. Solodiuk J.C., Scott-Sutherland J., Meyers M., Myetel et al. Validation of the Individualized Numeric Rating Scale (INRS): a pain assessment tool for nonverbal children with intellectual disability. *Pain* 2010; 150(2): 231-236. doi: 10.1016/j.pain.2010.03.016.
44. Gamble J.G., Rinsky L.A., Bleck E.E. Established hip dislocations in children with cerebral palsy. *Clin Orthop Relat Res* 1990; 253: 90-9.
45. Mubarak S.J., Valencia F.G., Wenger D.R. One-stage correction of the spastic dislocated hip. Use of pericapsular acetabuloplasty to improve coverage. *J Bone Joint Surg Am* 1992; 74(9): 1347-57.
46. Goodenough B., Addicoat L., Champion G.D. et al. Pain in 4- to 6-year-old children receiving intramuscular injections: a comparison of the Faces Pain Scale with other self-report and behavioral measures. *Clin J Pain* 1997; 13(1): 60-73. doi: 10.1097/00002508-199703000-00009.
47. Pritchett J.W. Treated and untreated unstable hips in severe cerebral palsy. *Dev Med Child Neurol* 1990; 32(1): 3-6. doi: 10.1111/j.1469-8749.1990.tb08460.x.
48. Abu-Saad H.H., Pool H., Tulkens B. Further validity testing of the Abu-Saad Paediatric Pain Assessment Tool. *J Adv Nurs* 1994; 19(6): 1063-71. doi: 10.1111/j.1365-2648.1994.tb01189.x.
49. von Baeyer C.L. Children's self-report of pain intensity: what we know, where we are headed. *Pain Res Manag* 2009; 14(1): 39-45. doi: 10.1155/2009/259759.
50. Cohen L.L., La Greca A.M., Blount R.L. et al. Introduction to special issue: Evidence-based assessment in pediatric psychology. *J Pediatr Psychol* 2008; 33(9): 911-5. doi: 10.1093/jpepsy/jsj115.
51. Jongudomkarn D., Angsupakorn N., Siripul P. The development and validation of the Khon Kaen University Pediatric Pain Assessment Tool for school-aged Isaan children in Thailand. *J Transcult Nurs* 2008; 19(3): 213-22. doi: 10.1177/1043659608317092.
52. Finley G.A., Kristjánsdóttir O., Forgeron P.A. Cultural influences on the assessment of children's pain. *Pain Res Manag* 2009; 14(1): 33-7. doi: 10.1155/2009/763031.
53. Mbada C.E., Orimolade A.E., Falujo O.C. et al. Development and preliminary testing of a culturally-friendly pain assessment tool for children (Crying Faces Pain Scale). *Med Rehabil* 2018; 22(3): 4-10. DOI: 10.5604/01.3001.0013.0209.
54. National Institute for Health and Care Excellence (NICE) guideline (2019). Cerebral palsy in adults: evidence reviews for identification of pain (NG119 Evidence reviews). <https://www.nice.org.uk/guidance/ng119/evidence/evidence-review-e-identifying-pain-pdf-239689926617>.
55. Jensen M.P., Engel J.M., McKearnan K.A., Hoffman A.J. Validity of pain intensity assessment in persons with cerebral palsy: a comparison of six scales. *J Pain* 2003; 4(2): 56-63. doi: 10.1054/jpai.2003.9.
56. Morkink L.B., Prinsen C.A., Bouter L.M. et al. The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) and how to select an outcome measurement instrument. *Braz J Phys Ther* 2016; 20(2): 105-13. doi: 10.1590/bjpt-rbf.2014.0143.

Address for correspondence

Adekola B. Ademoyegun
Department of Physiotherapy
Osun State University Teaching Hospital
PMB 5000, Osogbo, Nigeria
e-mail: aademoyegun@gmail.com