Chronic pain among children with cerebral palsy attending a Ugandan hospital: a cross-sectional study

Emma Nsalazi Bambi (emmanuelbambi@gmail.com)
Makerere University
Angelina Kakooza Mwesige
Makerere University
Hervé Monka Lekuya
Makerere University
Philip Kasirye
Makerere University
Richard Idro
Makerere University

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Abstract

Background: Children with cerebral palsy (CP) frequently experience chronic pain. The burden and severity of such pain is often underestimated in relation to their other impairments.Recognition and awareness of this chronic pain among children with CP constitute the cornerstone for caretakers and clinicians to improve the quality of life of those children. This study aimed to determine the prevalence of chronic pain among children with CP, and the factors associated.

Methods: A cross-sectional study of children with CP, aged 2 – 12 years, attending the CP rehabilitation clinic and Pediatric Neurology Clinic at Mulago Hospital, Uganda from November 2017 to May 2018. A detailed history and clinical examination were performed and the co-morbidities were determined. CP was classified using the Gross Motor Function Classification System (GMFCS), Manual Ability Classification System, Communication Function Classification System (CFCS), and the Eating and Drinking Ability Classification System (EDACS) and documented with the level of impairment in the different domains. Pain was assessed by using the revised Face, Legs, Activity, Consolability, Cry pain scale.

Results: A total of 224 children with CP were enrolled. The prevalence of chronic pain was 64.3%. The majority had spastic bilateral CP (77.8%), moderate pain lasting over 6 months, and none of them was on long-term pain management. Epilepsy (60.9%), behavioral problem (63.2%), hearing impairment (66,7%), learning problem (67,6%), dental caries (75%), gastro-esophageal reflux (75%), sleep disorders (79.5%), vision impairment (80%), and malnutrition (90%) were comorbid conditions of chronic pain in children with CP in this study. The factors independently associated with chronic pain among children with CP were the GMFC system level 4 & 5, CFCS level 4 & 5, EDACS level 4 & 5, female children, and caretaker aged more than 30 years.

Conclusion:

Two-thirds of children with CP attending rehabilitation in this hospital had chronic pain. None was receiving pain management. Chronic pain was associated with the presence of multiple co-morbidities and more severe disability. Rehabilitation and care programs for children with CP should include assessment of pain in routine care and provide interventions for pain relief in children with CP even at an early age.

Background

Cerebral palsy in children and adolescents is a major social disability, and affecting their quality of life, and largely of their caretakers. There is a growing recognition that pain is a significant problem in children and adolescents with cerebral palsy (CP) [1]. About 65 to 78% of children with CP are reported by their caretakers to experience pain [2]. Research has shown that children with CP who have pain participate less in everyday situations, experience lower quality of life than those without pain, and interferes with sleep [2]. Moreover, CP children who experience pain are more likely to struggle with anxiety, frustration, fear, depression, and behavior disorders such as aggression [3]. Self-report is

considered the gold standard for pain assessment due to pain's subjective and individual nature; this is particularly challenging for children with CP because some have a speech impairment, which makes it difficult to communicate their experience and others have such severe cognitive impairment that proxy reports are necessary [4]. Research supports this alternate approach to data collection or clinical assessment, as Engel and colleagues found the agreement between parent and child report of pain occurrence was 93% [2]. Currently, there is very little literatures about how common is chronic pain among children with CP in sub-Saharan African, especially in Uganda, where there is a high prevalence of CP among children resulting from several peri-natal causes. There is little knowledge about the factors that predispose to this pain and their associated complications. Recognition and awareness of chronic pain and the burden of associated factors of this pain among children with CP could help us to provide an accurate assessment, and plan for better interventions to improve the quality of life of those children. The study aimed to determine the prevalence of chronic pain in children with CP, to describe the severity and comorbidities of pain, and determine the factors associated with such pain.

Methods

Study design and setting

This cross-sectional study was carried out in the Cerebral Palsy Rehabilitation Clinic and the Pediatric Neurology Clinic at Mulago National Referral Hospital, Kampala, Uganda, between November 2017 and May 2018.

Study participants

Cerebral palsy is a neuro-developmental condition comprising a group of permanent disorders of movement and posture that are attributed to non-progressive injury that occurred in the developing fetal or infant brain.

Study participants were Children of 2–12 years old with a documented diagnosis of CP, attending regularly the above-mentioned clinics for their follow-up, accompanied by a caretaker. The latter was responsible for providing most of the material and emotional requirements to the child for at least 3 months.

Only patients whose parents or caregivers provide informed consent were included.
Participants who came with caregivers who were unable to provide adequate information about the child on the study or refuse to provide informed consent were excluded in the study.

Study procedure

The study team consisted of medical personnel working within the above-mentioned Paediatrics clinics with additionally trained research assistants to do the enrolment, consent, history, and physical examination.

The detailed clinical history as well as comorbidities were obtained from the parents or caretakers; the child was examined to obtain the features and complications of CP. The patient's level of physical functioning was assessed using the Gross Motor Function Classification system (GMFCS). The GMFCS has levels I-V with level I indicating a good level of functioning while level V is the worst depicting severe handicap. The ability of hand functioning was also assessed using the manual ability classification system (MACS) which has also levels I-V. This was followed by assessing the child's communication ability using the communication function classification system (CFCS). The eating and drinking ability classification system ((EADACS) was assessed as well, knowing that EADACS has levels I-V.

Measurement of pain in children with CP

Pain was assessed using the revised face, legs, activity, cry and consolability (rFLACC) scale for children aged between 2–12 years old. The rFLACC scale questionnaires for children with CP were filled to determine the pain and the severity of pain. This instrument has been recognized to be reproducible, valid, and reliable in assessing pain in children with CP not able to self-report pain. The internal consistency is excellent with a Cronbach's alpha of 0.9023 and 0.9753 (two raters). A test–retest showed excellent intra-rater reliability with an intra-class correlation (ICC) of 0.97530.

Data analysis

Descriptive statistics were used to explore baseline characteristics of the participants and caretakers; they were presented as means and medians, ranges, and IQR. Variables whose p-value of the Unadjusted OR (bivariable level) was less than 0.2 were considered at multivariable logistic regression analysis to find out factors that significantly determine having chronic pain among children with CP. Significance was set at a p-value of ≤ 0.05.

Ethical approval: Approval was sought from the Department of Paediatrics and Child Health of Makerere University College of Health Sciences, as well as the School of Medicine Research and Ethics Committee (SOMREC).

Table 1
Socio-demographic and medical history of children with CP

Characteristics	Frequency (N = 224)	Percentage (%)
Age (in years)	3.6 (2.5–5.6)	-
Median (IQR)		
Sex	140	62.5
Male	84	37.5
Female		
Pregnancy/Delivery	123	54.9
Normal	84	37.5
Difficulty	17	7.6
Not known		
Delivery	190	84.8
Term	9	4.0
Preterm	25	11.2
Not known		
Birth weight (in kilograms)	3.3 (3.1,3.4)	-
Mean(SD)		
Neonatal problems	178	79.5
Yes	46	20.5
No		

Table 2
Socio-demographic characteristics of the main caretakers of children with CP

Characteristics	Frequency	Percentage (%)
Age (in years)	30 (27,38)	
Median (IQR)		
Sex	22	9.8
Male	202	90.2
Female		
Relationship to child	179	79.9
Mother	18	8.0
Father	27	12.1
Others		
Employment status	85	37.9
Employed	139	62.1
Unemployed or others		
Education status	18	8.0
No schooling	80	35.7
Primary	87	38.9
Secondary	39	17.4
Tertiary		
Marital status	163	72.7
Married	27	12.1
Single	34	15.2
Other*		
Religion	183	81.7
Christian	41	18.3
Muslim		

**includes separated, divorced and widowed*

Characteristics	Frequency	Percentage (%)
Family size	91	40.6
< 4 members	133	59.4
≥ 4 members		
**includes separated, divorced and widowed*		

Table 3
Chronic pain according to clinical characteristics of children with CP

Characteristics	Frequency (N = 224)	Percentage (%)
Chronic Pain		
Yes	144	64.3
No	80	35.7
Chronic pain according to the type of CP		
Spastic bilateral	91/117	77.8
Spastic unilateral	12/24	50.0
Dyskinetic CP	27/55	49.1
Ataxic CP	4/7	57.1
Non classifiable	10/21	47.6
Chronic pain according to GMFC		
Level 1	1/14	7.1
Level 2	2/14	14.3
Level 3	5/13	38.5
Level 4	39/65	60.0
Level 5	97/118	82.2
Chronic pain according to MACS		
Level 1	4/27	14.8
Level 2	7/27	25.9
Level 3	10/19	52.6
Level 4	29/39	74.4
Level 5	94/112	83.9
Chronic pain according to CFCS		
Level 1	0/19	0
Level 2	10/30	33.3
Level 3	14/29	48.3
Level 4	26/35	74.3
Level 5	94/111	84.7

Characteristics	Frequency (N = 224)	Percentage (%)
Chronic pain according to EADACS		
Level 1	8/31	25.8
Level 2	9/35	25.9
Level 3	30/47	63.8
Level 4	81/95	85.3
Level 5	16/16	100

Table 4
Severity and duration of pain among children with CP

Severity of pain	Duration of pain- n (%)	
	3–6 months (n = 33)	More than 6 months (n = 111)
Moderate pain	28 (84.8)	64 (57.7)
Severe pain	5 (15.2)	47 (42.3)

Table 5
Comorbid conditions among children with CP

Comorbidity	Chronic pain/ N	Prevalence (%)	95%CI
Illnesses	84/138	60.9	48.6–75.4
Epilepsy	31/39	79.5	54.0-100
Sleeping disorders	16/20	80.0	45.7–100
Impairment vision	12/16	75.0	38.8–100
Dental caries	10/11	90.9	43.6–100
Malnutrition	6/8	75.0	27.5–100
Gastro-esophageal reflux disease	4/6	66.7	18.2–100
Impairment hearing			
Behavioral problem	55/87	63.2	47.6–82.3
Presence of aggressive behavior	48//103	46.6	34.4–61.8
Plays with other children			
Learning problem			
Schooling	2/14	14.3	1.7–51.6
Not at School	142/210	67.6	57.0-79.7

Table 6
Factors independently associated with chronic pain among children with CP

Characteristics	Overall N = 224	Unadjusted OR (95% CI)	p-value	Adjusted OR (95%CI)	p-value
Child's gender					
Male	140(62.5)	1.00	1.00		
Female	84(37.5)	1.67(0.93–2.98)	0.085	2.54(1.13–5.73)	**0.024**
CP classification					
Spastic bilateral	117(52.2)	1.00		1.00	
Spastic unilateral	24(10.7)	0.28(0.12–0.71)	0.007	0.80(0.25–2.53)	0.701
Dyskinetic CP	55(24.6)	0.27(0.14–0.55)	0.001	0.64(0.27–1.56)	0.328
Ataxic CP	7(3.1)	0.38(0.08–1.81)	0.225	1.45(0.15–13.71)	0.747
Non-classifiable	21(9.4)	0.26(0.10–0.68)	0.006	0.75(0.18–3.02)	0.681
Classification					
GMFCS					
Level 1, 2, & 3	41(18.3)	1.00		1.00	
Level 4 & 5	183(81.7)	11.94(5.14–27.7)	< 0.001	3.75(1.38–10.20)	**0.009**
MACS					
Level 1, 2, & 3	73(32.6)	1.00		1.00	
Level 4 & 5	151(67.4)	10.9(5.7–20.9)	< 0.001	2.54(0.99–6.5)	0.052
CFCS					
Level 1, 2, & 3	78(34.8)	1.00		1.00	
Level 4 & 5	146(65.2)	10.4(5.5–19.7)	< 0.001	2.46(1.03–5.90)	**0.044**
EADACS					
Level 1, 2, & 3	113(50.5)	1.00		1.00	
Level 4& 5	111(49.5)	9.73(4.96–19.09)	< 0.001	3.32(1.37–8.04)	**0.008**
Comorbidities					
No	60(26.8)	1.00			

Characteristics	Overall N = 224	Unadjusted OR (95% CI)	p-value	Adjusted OR (95%CI)	p-value
Yes	164(73.2)	0.96(0.52–1.78)	0.893		
Main caregiver's age					
< 30	104(46.4)	1.00		1.00	
30+	120(53.6)	0.62(0.35–1.07)	0.087	0.45(0.21–0.95)	**0.035**

Results

Between November 2017 and May 2018, a total of 234 children with CP attended the Cerebral Palsy Rehabilitation Clinic or General Paediatric Neurology Clinics in Mulago Hospital and were invited to participate in the study; 4 refused consent, 2 were outside the required age range of 2-12 years, 4 were unable to participate due to other reasons and 224 were recruited. The majority of participants were male (62.5%), with an overall median age of 3.6 years (Table 1). Most of the caretakers were literate married Christian mothers, with no formal employment, living in a family size of more than 4 people (Table 2). One hundred forty-four (144) of the 224 children presented with chronic pain giving a prevalence of 64.3% (Table 3). The highest percentage of children with chronic pain was observed with children with spastic bilateral CP (77.8%). None of those children with CP in chronic pain was on long-term pain management. Participants in level 4 & 5 of GMFCS, MACS, CFCS, and EADCS had a high prevalence of chronic pain compare to participants in level 1, 2 & 3 (Table 3). The majority of participants had moderate pain, but the duration of pain was more than 6 months (Table 4). Epilepsy, sleep disorders, impaired vision, impaired hearing, dental caries, malnutrition, gastroesophageal reflux, behavioral problem, and earing problem were comorbid conditions in this study (Table 5). Female children had at least two times the odds of having chronic pain compared to male children (OR, 95% CI: 2.50, 1.13-5.73, p=0.024). Also, GMFC classification was significantly associated with having chronic pain in this population of children with CP. Children who were in level 4 & 5 had almost 4 times the odds of having chronic pain compared to those that were in either level 1 or 2 or 3; {OR, 95% CI: 3.75 (1.38-10.20), p=0.009}. Also, EADAC and CFC levels determined the risk of having chronic pain where those in level 4 or 5 had a higher risk of having chronic pain compared to those in level 1 or 2 or 3 {OR, 95% CI: 2.46 (1.03-5.90), p=0.044 and 3.32, 1.37-8.04; p=0.008 respectively}. The results also further revealed that children with older caretakers (30+ years) were less likely to have chronic pain compared to those taken care of by caretakers less than thirty years {OR, 95%CI: 0.45 (0.21-0.95), p=0.03} (Table 6).

Discussion

This study set out to determine the prevalence of chronic pain and associated factors among children with CP. We found that 64.3 % of these patients who had chronic pain with severe gross motor function level involvement or disability were more at risk. The prevalence of chronic pain in this study is consistent

with previous studies done in Malaysia in 2015 [5] and turkey in 2017 [6], where the prevalence of chronic pain in children with CP in both 2 studies was found to be 65%. Also, the prevalence of pain in children with CP in this study is similar to a previous report that indicated that around 60 % of children with CP experience recurrent pain on a daily or weekly basis that significantly interferes with their daily activities [7]. In this study, the most common type of CP with the highest prevalence of chronic pain was spastic bilateral CP. Knowing that children with spastic bilateral CP have more severe functional motor limitation and experiencing stiffness and contractures which usually prone them to ongoing pain. A similar pattern was found in a study done by Badia *et al.* in 2014 where most participants with pain had bilateral spastic [1]. Participants with GMFCS, MACS, CFCS, EADACS level IV, V had the highest prevalence of pain in this study; these findings are concurring with a previous cross-sectional multi-centre European study done by Gibson *et al.* in 2009 that reported a higher frequency and severity of pain commonly found if the child is more severely impaired in terms of walking ability, hand function, feeding and communication difficulties, intellect and CP type [2]. Another study done in Sweden in 2017 by Lena *et al* indicated that children with Level V in both Gross Motor Function Classification System (GMFCS) and Manual Ability Classification System (MACS) were associated with the highest prevalence of pain [8]. Furthermore, 2 other studies done by Parkinson *et al.in* 2013 [9] and Barney *et al* in 2013 [10] discovered that pain frequency, intensity, and duration were significantly predicted by GMFCS level, being greater among children with a higher GMFCS level 4,5. Similarly, a study done in Turkey in 2017 [6], showed that children with GMFCS 4,5 had a high percentage of getting pain compared to patients at GMFCS levels 1, 2, and 3. In this study majority of participants had moderate pain. A similar pattern was reported in a study done in Malaysia by Subhashini *et al.* in 2015 where patients with moderate pain were more than those with severe pain [5]. These findings are also similar to a previous study done in Spain that showed that persons with CP experience recurrent pain of moderate-intensity on a daily or weekly basis that significantly interferes with daily activities [7].

Epilepsy, sleep disorders, vision impairment, hearing impairment, dental caries, malnutrition, gastro-esophageal reflux disease, behavioral problem, and learning problem were comorbid conditions in children with pain and CP in this study. These findings are similar to a previous study done by Badia *et al.* in 2014, where epilepsy, hearing disabilities, visual disability, behavior problem, communication problem, feeding problem were the most common comorbid conditions in children with pain and CP [1]. Another study done by Raymond *et al.* presented similar findings to ours where the commonest comorbid conditions that parents reported in their children with pain and CP were a developmental delay, vision problem, learning problem, behavior problem, speech problem, seizure disorder, hearing impairment, and sleep disturbance [11]. In this study, we found that gross motor function classification level 4 & 5, Communication function classification level 4 & 5, eating and drinking ability classification system level 4 & 5, female participants, and caretaker age of more than 30 years were independently significant factors associated with chronic pain. Indeed in the multi-centre European cross-sectional study done by Parkinson *et al.*in 2009, it was reported that a higher frequency and severity of pain was common if the child was more severely impaired in terms of walking ability, hand function, feeding and communication difficulties, intellect and CP type [2]. Knowing that patients with GMFCS, EADACS, CFCS level 4 & 5

usually have a severe disability in term of motor impairment and persons with severe impairment tend to have more spasticity, which causes ongoing pain. Also in this study, we found that female participants had at least two times the odds of having chronic pain compared to the male child. A similar pattern was reported by Christine et al, in 2004 where the female was reported to have a greater frequency of pain compare to the male [12]. Children with older caretakers (30+ years) were less likely to have chronic pain. This may be explained by the fact that older caretakers have much more experience of recognition of pain, and may be giving more support to relieve this chronic pain.

The limitation of this study was a relatively small sample size. The nature of the study design could only describe chronic pain among children with CP. The type and location of pain could not be clearly ascertained when dealing with children with speech and cognitive impairments. Although we used validated tools from previous studies, children with CP in sub-Saharan African countries could have benefited from an adjusted tool regarding local realities.

Conclusions

Approximately two-thirds of children with CP attending rehabilitation in this tertiary hospital experience chronic pain. The majority of these children have moderate pain without long-term pain management. Comorbid conditions such as epilepsy, sleep disorders, impairment of vision, impairment of hearing, dental caries, malnutrition, gastroesophageal reflux, behavioral problem, and learning problem are associated with chronic pain among children with CP. Severe disability in terms of GMFC, CFCS, EADACS (level 4 & 5), and female participants were associated with chronic pain among children with CP. Children with older caretakers (30 years and above) may likely experience less chronic pain. We recommend rehabilitation and care programs for children with CP to assess chronic pain in routine care and provide interventions for pain relief in children with CP even at an early age.

List Of Abbreviations

CP: Cerebral palsy; CFCS: Communication function classification system; EADACS: Eating and drinking ability classification system; GMFCS: Gross- motor function classification system; MACS: Manual ability classification system; rFLACC: Revised face, legs, activity, consolability, cry pain scale

Declarations

Ethics approval and consent to participate, and for publication: This study was reviewed and approved by the Makerere University School of Medicine Research and Ethics Committee. Written informed consent was obtained from the parent/guardian in order to participate in the study. All study methods were performed in accordance with the relevant guidelines and regulations of Declaration of Helsinki.

Availability of dataset and material: The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Competing interests: The authors declare that they have no competing interests.

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Authors' contributions: ENB, AK, HML, KP & RI

ENB conceptualized the study, designed the methodology, supervised data collection, and was the major contributor to the manuscript; AK conceptualized the study, contributed to designing of the methodology, and manuscript writing; HML contributed to the data collection, reviewed study data, and contributed to manuscript writing; KP contributed to study methodology and manuscript writing; RI contributed to the methodology, manuscript writing and critical review. All authors read and approved the final manuscript.

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