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Post-Operative Pain Assessment and Management in Cerebral Palsy (CP): A Two-Pronged Comparative Study on the Experience of Surgical Patients

Nanfang Xu [a,*], Hiroko Matsumoto [b], David Roye [b], Joshua Hyman [b]

[a] Department of Orthopaedics, Peking University Third Hospital, Beijing, China
[b] Department of Orthopaedic Surgery, Columbia University Medical Center, New York, NY, United States of America

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ABSTRACT

Introduction: This study compares the current practice patterns of pain assessment and management between children with and without CP following either posterior spinal instrumentation and fusion (PSIF) or hip osteotomy (HO).
Methods: Two cohorts of CP patients were retrospectively identified and matched with non-CP patients based on age, surgical procedure, and approach to post-operative pain management. Sixteen CP patients undergoing PSIF and twenty-two undergoing HO were respectively matched with the same numbers of non-CP patients receiving the same procedures. The frequency of assessments conducted, highest pain scores recorded on each post-operative day (POD), and the amount of adjuvant analgesics administered were collected for POD 0–4.
Results: Patients with CP were significantly more frequently evaluated for pain post-operatively, tended to have lower pain scores as measured by current scales, and received slightly fewer analgesics. Patients with CP differed from their non-CP counterparts in both frequency and method of post-operative pain assessment.
Conclusions: The purpose of this study is to elucidate the current state of post-operative pain assessment and management in children with CP undergoing major orthopaedic surgeries, to improve CP patient/caregiver understanding and expectation of the post-operative experience regarding pain, and to provide recommendations for improving the post-operative care for these patients.

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Introduction

CP is a clinical diagnosis defined as a non-progressive cerebral insult occurring in the developing fetal or infant brain. It can result in a spectrum of permanent musculoskeletal and movement disorders (Rosenbaum et al., 2007). In addition to the characteristic involuntary muscle contractions, these patients suffer from a number of comorbid conditions, including postural difficulties that negatively affect feeding (Charpentier, Morgan, & Harding, 2018), impairments of vision and hearing, speech disorders, oral-motor dysfunction, epilepsy, and chronic pain (Venkateswaran & Shevell, 2008). Among these, pain, with reported prevalence rates approaching 60% (Engel, Kartin, & Jensen, 2002; Engel, Petrina, Dudgeon, & McKearnan, 2005; Ostojic, Paget, & Morrow, 2018; Ramstad, Jahnsen, Skjeldal, & Diseth, 2011), has been thought to result from various etiologies, including soft tissue strain, gastrointestinal and genitourinary disturbances, and aberrant biomechanical forces due to spasticity (Engel et al., 2005; Dudgeon et al., 2005; Oberlander, O'Donnell, & Montgomery, 1999). While the particular pathophysiology has not been described, psychological distress, sleep disturbance, and poor self-image are well known consequences of inadequate pain management in these patients (McKearnan, Kieckhefer, Engel, Jensen, & Labyak, 2004; Tervo, Symons, Stout, & Novacheck, 2006).

Pain has been described by the International Association for the Study of Pain as an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage. The effective evaluation and treatment of pain is complicated by its subjective nature, as a result of which self-report has become the gold standard (Drendel, Kelly, & Ali, 2011). However, for the estimated 23–44% of individuals with CP who are cognitively-impaired, and the 42–81% who have speech impairment, their ability to participate in the self-report of pain may be limited or impossible (Odding, Roebroeck, & Stam, 2006). The literature discussing the pain experience in patients with CP, possible contributors to pain, and the various barriers to adequate pain assessment has grown over the past decade (Ghai, Makkar, & Wig, 2008; Hadden & von Baeyer, 2005; Houlihan, O'Donnell, Conaway, & Stevenson, 2004; Hunt & Franck, 2011; Lauder & White, 2005; Nolan, Chalkiadis, Low, Olesch, & Brown, 2000; Russo, Miller, Haan, Cameron, & Crotty, 2008; Swiggum, Hamilton, Gleeson, Roddey, & Mitchell, 2010; Terstegen, Koot, de Boer, & Tibboel, 2003).

* Corresponding author.
E-mail addresses: xunanfang@foxmail.com (N. Xu), jh736@cumc.columbia.edu (J. Hyman).

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Although self-report is the gold standard in pain measurement, few CP patients are capable of self-report. As a result, observational measures have been used in the study of postoperative pain in this population (Breau & Burkitt, 2009). Tools commonly used in children and in individuals who are unable to verbalize pain include the Faces Legs Activity Cry Consolability (FLACC) behavioral scale, the FACES scale, and the numerical rating scale (NRS). However, none has been specifically validated for use in the population with CP (Voepel-Lewis, Merkel, Tait, Trzcinka, & Malviya, 2002). Furthermore, the FLACC, FACES, and NRS are frequently combined in studies since they all use a 0–10 scale. However, there is inconsistent evidence regarding the correlation between the three different instruments (Hadden, LeFort, O'Brien, Coyte, & Guerriere, 2015; Hicks, von Baeyer, Spafford, van Korlaar, & Goodenough, 2001; Massaro et al., 2014; Moore, Wester, Sunder, Schrock, & Park, 2013; Voepel-Lewis, Zanotti, Dammeyer, & Merkel, 2010). Typically, correlations of ≥0.6 are considered good to excellent associations. In one study, the FLACC and FACES instruments had a correlation of 0.58 in the entire patient population (aged 3–7), and this correlation was increased to 0.83 when data was analyzed for patients older than 5 (Willis, Merkel, Voepel-Lewis, & Malviya, 2003). However, in another study, the FLACC and FACES instruments only had a correlation of 0.35 in communicative children aged 5–16 (Nilsson, Finnstrom, & Kokinsky, 2008). While other pain instruments such as the Revised FLACC and Non-Communicating Children's Pain Checklist (NCCPC) have been developed to address this disparity, they are rarely employed in the hospital setting (Barbi, Massaro, & Badina, 2011; Breau, 2003; Malviya, Voepel-Lewis, Burke, Merkel, & Tait, 2006).

Inadequate assessment may translate to unsuccessful treatment of pain in CP patients. A study evaluating postoperative pain management showed that children with cognitive impairment were assessed for pain less frequently, and received less total opioid doses than children without cognitive impairment (Malviya et al., 2001). Apart from difficulties in assessing pain, CP patients are perceived to be particularly susceptible to central nervous system and respiratory depression, which may influence providers' decisions with regard to the use of opioids.

The prevalence of CP is about 2.0 per 1000 births, and approximately 60% of CP patients will require an orthopaedic procedure by the age of 8 (Chicoine, Park, & Kaufman, 1997; Odding et al., 2006). Spinal deformity correction (posterior spinal instrumentation and fusion, PSIF) and hip osteotomy (HO) represent two common procedures that are frequently associated with significant postoperative pain (Shrader et al., 2015). Despite the recent interest in the study of pain in cognitively-impaired individuals, questions remain regarding the assessment and treatment of pain in CP patients (Shaikh & Hegade, 2017).

Materials and methods

After obtaining Institutional Review Board approval, a two-pronged retrospective case-control study was performed, using Current Procedural Terminology (CPT) codes to identify patients undergoing PSIF or HO between 2008 and 2016. All procedures were performed by a single group of pediatric orthopaedic surgeons at a tertiary care children's hospital. The participants had to meet the following inclusion criteria: (1) Patient age 5–21 years old at the time of the surgery, (2) the CP cohorts were identified based on ICD-9 codes (and further confirmed by review of provider-patient encounter documentation), and (3) the control cohorts were selected based on similar routes of postoperative analgesia and closest age match. Exclusion criteria were: (1) incomplete pain data or medication dose history during inpatient stay, (2) intubation in the pediatric intensive care unit (PICU) for >48 h postoperatively, (3) documentation of neuromuscular disease and/or developmental delay in the non-CP cohorts.

Data were collected for pertinent demographic information, previous medical history, length of stay in the hospital, and surgical details. The FLACC scale was used in patients who could not communicate, while the FACES or the NRS was used in patients who were able to communicate their pain. Postoperative data were collected for POD 1–4, with the date of surgery defined as POD 0. Descriptive analyses were conducted to determine: Total number of pain assessments, highest pain score per day, the total amount of opioids given (normalized to morphine equivalents by weight, MEq/kg), and the method of administration (continuous infusion, patient-controlled analgesia (PCA), clinician bolus, and oral dose). Student's *t*-tests were used to compare differences in pain assessment frequency, pain score, and total MEq/kg between the CP and non-CP cohorts. Statistical significance was defined as $P < 0.05$.

Results

PSIF branch

This branch of retrospective study identified 16 patients with the diagnosis of CP who were age matched to 16 children with adolescent idiopathic scoliosis (AIS). Demographic data were presented in Table 1. The CP group was predominantly male, and most had a prior history of surgery (e.g. tendon release, placement of gastrostomy tube, tracheostomy, etc.). On average, CP patients had a greater number of levels fused and were hospitalized for a longer period of time following surgery.

All patients in the AIS group were able to self-report pain using the NRS scale. Only one individual with AIS was initially assessed with the FLACC scale in the PICU following surgery; this patient reported pain using the NRS scale following extubation. On the other hand, six children (37.5%) with CP were able to self-report pain using the NRS scale, and one (6.3%) was assessed using the FACES scale. The other nine (56.3%) were unable to self-report pain, and were assigned pain scores using the FLACC scale throughout the recorded assessment period.

The average number of documented pain assessments during POD 1–4 for CP patients (56 recorded assessments) was significantly higher ($P = 0.009$) than for AIS patients (36 recorded assessments) (Table 2). All individuals stayed in the hospital through POD 4, and all had at least one recorded pain assessment on each POD. The highest pain scores were averaged for each group on each POD (Table 2). Scores for the AIS group remained higher than those for the CP group throughout the assessment period (Fig. 1). The highest overall pain score recorded for the assessment period was also higher in AIS patients (8.8) than in CP patients (5.9) (Table 2). No difference in pain assessment, pain score or MEq/kg dose of opioids was found between the 7 patients

Table 1
Patient demographics and surgical details.

	PSIF branch		HO branch	
	CP	Non-CP	CP	Non-CP
Sex				
Male	11	3	16	11
Female	5	13	6	11
Age at surgery	14.8 ± 2.04	14.5 ± 1.99	10.7 ± 4.7	8.7 ± 5
Length of surgery (minutes)	457 ± 115	466 ± 82	229.9 ± 70.5	223.9 ± 94.7
Number of levels fusioned	17 ± 0.6	13 ± 1	N/A	N/A
Type of osteotomy				
Femur	N/A	N/A	8	11
Pelvis	N/A	N/A	4	6
Femur + pelvis	N/A	N/A	10	5
Length of stay	9.4 ± 1.9	5.9 ± 0.9	5.1 ± 1.4	4.5 ± 1.3
Previous surgeries				
No	3 (18.8)	15 (93.8)	0	18 (81.8%)
Yes	13 (81.3)	1 (6.3)	22 (100%)	4 (18.2%)

Table 2
Number of recorded pain assessments and highest recorded pain score in the PSIF branch.

	CP (n = 16)	Non-CP (n = 16)	P value (if significant)
Number of recorded pain assessments			
POD 1	20.6	17.0	
POD 2	17.0	8.9	$P = 0.001$
POD 3	11.8	5.9	$P = 0.006$
POD 4	6.8	4.3	
POD 1–4	56.1	36.0	$P = 0.002$
Highest recorded pain score			
POD 1	3.7	7.1	$P = 0.001$
POD 2	4.2	5.9	
POD 3	3.8	5.0	
POD 4	3.0	5.1	
Highest overall pain score recorded during POD 1–4	5.9	8.4	

Table 3
Number of recorded pain assessments and highest recorded pain score in the HO branch.

	CP (n = 22)	Non-CP (n = 22)	P value (if significant)
Number of recorded pain assessments			
POD 1	7.7	4.5	
POD 2	7.6	3.6	$P = 0.007$
POD 3	4.9	3.6	
POD 4	3.9	3.1	
POD 1–4	28.8	19.6	$P = 0.034$
Highest recorded pain score			
POD 1	2.2	3.6	
POD 2	2.3	2.5	
POD 3	2.2	2.5	
POD 4	1.9	1.8	
Highest overall pain score recorded during POD 1–4	5.2	6.0	

with CP who were able to use the self-report pain scales and the 9 who were assessed using the FLACC scale.

While intubated, patients received a continuous infusion of IV fentanyl as a sedative-narcotic. They were then transitioned to either IV morphine or IV hydromorphone by continuous infusion or via PCA, where patients were allowed to request narcotic as needed with a predefined hourly lockout period. Ninety-four percent of children with CP were given continuous infusion only, and one was prescribed a combination of PCA and continuous infusion. AIS patients were primarily prescribed a combination of PCA and continuous infusion (87.5%), while two (12.5%) were given PCA alone. All patients received additional clinician boluses for breakthrough pain. All AIS patients and 80% of CP patients were transitioned to oral (PO) opioids by POD 4.

Overall, there was no significant difference between the total MEq/kg given over POD 1–4 (CP 4.5 vs. AIS 4.2) (Table 3). CP patients received more MEq/kg on POD 1 (2.5 vs. 1.5, $P = 0.031$) and less MEq/kg on POD 2 (0.5 vs. 1.0, $P = 0.008$) and POD 3 (0.4 vs. 0.7, $P = 0.009$) (Fig. 2). The precipitous drop in MEq/kg seen between POD 1 and 2 corresponds with the average time of extubation (POD 1.2) in the CP group.

Hip osteotomy branch

There were 22 CP patients who had HO and they were matched to non-CP HO patients with the same postoperative analgesic modalities and closest age (Table 1). The CP group had a higher percentage of males compared to the non-CP group (72.7% vs. 50%). All CP patients had a history of prior surgery, whereas 82% of patients in the non-CP group also had a history of prior surgery. On average, CP patients remained hospitalized for a longer period of time following surgery.

The CP group was assessed for pain more frequently on each POD. This difference was statistically significant on POD 2 (7.6 vs. 3.6, $P = 0.007$) and throughout POD 0–4 when all pain assessments were summed (28.8 vs. 19.6, $P = 0.034$) (Table 3). The pain scores were higher in the non-CP group on each POD, but none of these differences were statistically significant. As in the PSIF branch of the study, the majority of CP patients were unable to self-report pain and were assessed using the FLACC scale, while almost all non-CP group children were able to self-report pain using the NRS or the FACES scale.

As patients transitioned to oral pain medications, the CP group was given a higher amount of toradol/kg on POD 3 (0.98 vs. 0.45, $P = 0.047$). Similarly, CP patients received more acetaminophen throughout POD 1–4, although the amount was not significantly different between the two groups. Subgroup analysis based on modality of postoperative pain management did not have enough power to provide statistical significance, but was useful to highlight trends in this study population. Overall, CP patients received less total MEq/kg during POD 0–4 than non-CP patients (2.0 ± 1.0 vs. 2.3 ± 1.3), but again, the difference was not significant statistically.

Discussion

The management of postoperative pain in CP patients is complicated by a number of factors. This study sought to investigate the differences in pain assessment and treatment in children with and without CP during the immediate postoperative period. Our data showed that CP patients were assessed more frequently than their peers, and their lower pain scores indicated that they were perceived to be experiencing less pain. However, the issue of particular gravity is whether or not pain was being properly assessed in these patients. Lastly, there was no

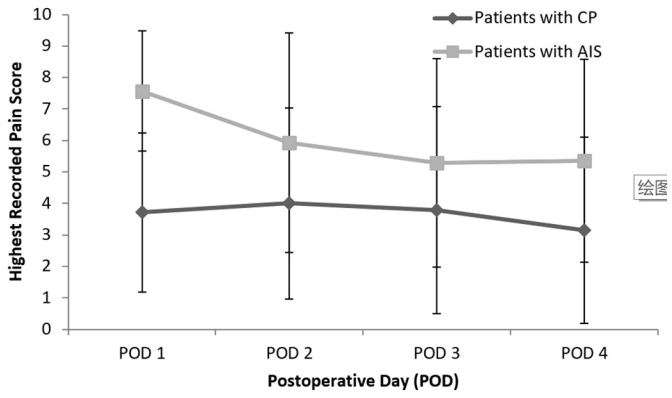

Fig. 1. Average highest daily pain score recorded for the PSIF branch. *Denotes $P < 0.05$.

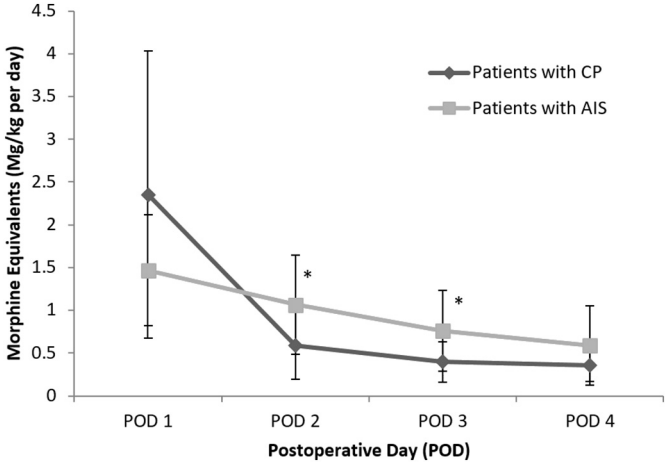

Fig. 2. Average MEq/kg received in the PSIF branch on postoperative days 1–4. *Denotes $P < 0.05$.

statistical difference between the amounts of overall analgesics received by the CP and the non-CP groups.

Our data reporting the frequency of pain assessment differed from a previous study, which found children with cognitive impairment received less frequent pain assessment on POD 1–3 (Malviya et al., 2001). The observation of increased frequency of pain assessments was consistent with the notion that CP patients often require increased level of nursing care, and may be explained in part by the standard-of-care practice at this institution to record hourly pain assessments while patients are being managed in the PICU. As an example, while the difference between the number of assessments in the CP and the AIS groups was not significant on POD 1 (20.6 vs 17.0), the number of assessments reached significance on POD 2. CP patients often remained intubated postoperatively. They were managed in the PICU longer than the AIS group before being transferred to wards, where hourly assessments were no longer standard-of-care. Therefore, the observation that their pain was assessed more frequently early in the postoperative course was expected. However, children with CP continued to be assessed more frequently than the AIS group by POD 3 (11.8 vs 5.9), suggesting that providers remained attentive to this population, despite the inherent difficulties in assessing their pain. This also suggests that the lower pain scores and lower MEq/kg received by the CP group were not the result of patient neglect.

Despite the higher frequency of assessments, whether or not pain in children with CP and cognitive impairment is being adequately assessed by existing instruments remains an issue of debate (Ghai et al., 2008; Voepel-Lewis et al., 2002). We observed that pain scores tended to be lower in children with CP, which was similar to findings in children with cognitive impairment (Malviya et al., 2001). Over the past decade, there has been an increased interest in the assessment of pain in the cognitively-impaired, and several instruments have been investigated for their utility in this population. The NCCPC – Postoperative Version (NCCPC-PV) is designed for use in severely-impaired children, but is limited by the extensive assessment requiring description of 27 behavior types during a 10 min observation period (Breau, Finley, McGrath, & Camfield, 2002). The FLACC scale has shown good reliability and validity, but low agreement in some categories (Merkel, Voepel-Lewis, Shayevitz, & Malviya, 1997). Issues facing effecting observational assessment with the FLACC or NCCPC scale include the presence of atypical facial responses, motor impairment, and other idiosyncratic behaviors in this population that could be mistaken for pain, resulting in inaccurate pain assessment (Breau & Burkitt, 2009). Finally, the Revised FLACC has attempted to account for some of the atypical behaviors which have been noted in cognitively-impaired individuals, particularly in those with spasticity (Breau & Burkitt, 2009; Malviya et al., 2006). As more than half of the sample used in the validation study for the Revised FLACC was diagnosed with CP, and the presence and distribution of spasticity were included in the analysis of reliability, this tool appears particularly applicable to non-communicating CP patients. However, while these tools may exist, the likelihood that they are being used is limited by the difficulties which face the introduction of any new instruments into the clinical environment, including staff acceptance, ease of use, and institutional protocols, etc. (Barbi et al., 2011; Hunt & Franck, 2011). Additionally, given the high variability of physical and cognitive involvement which characterizes CP, instruments designed to assess patients with cognitive impairment which ignore the specific neuromuscular manifestations of CP may be inadequate (Malviya et al., 2006).

Several other groups have also developed pain instruments for individuals with cognitive impairments. Collignon et al. made a 22-item pain-assessment instrument for patients with multiple handicaps. However, when it was validated, the study included both adults and children. This was beneficial for pain assessment over a lifespan, but weakened its validity in the immediate postoperative period for children (Collignon & Giusiano, 2001). Boldingh et al. developed a pain assessment tool for the CP population, but it required a minimum score of 25 on the Columbia Mental Maturity Scale (Boldingh, Jacobs-van der Bruggen, Lankhorst, & Bouter, 2004). Researchers at Wisconsin Children's Hospital also created a pain scale, but the number of classifications limited the precision of its assessment (Soetenga, Frank, & Pellino, 1999). The Nursing Assessment Pain Intensity (NAPI) scale was praised for its high degree of clinical utility. However, CP patients comprised <10% of the sample in the validation study (Schade, Joyce, Gerkensmeyer, & Keck, 1996).

Although not statistically significant, the lower MEq/kg received by the CP groups (throughout POD 2–3 in the PSIF branch and POD 1–4 in the HO branch) appeared to correspond to their lower pain scores, suggesting that they were given opioid and adjuvant pain medications according to the observed experience of pain. Indeed, it has been hypothesized that individuals with severe neurological impairment may have a blunted pain response and experience pain differently from the rest of the population (Oberlander et al., 1999). However, these observations were limited by the notion that pain might not be accurately assessed in these patients. Our ability to make definitive conclusions is limited by the study design and the various challenges intrinsic to retrospective chart review studies, including the inaccuracy of available data and sporadic recording of the response to pain interventions. There were additional difficulties in accurately assessing cognitive status or ascribing an IQ to patients, who were assessed using the NRS or FACES scales, to determine if they were cognitively capable of giving reliable self-report.

The current study revealed some of the difficulties in the assessment of pain in non-communicative CP patients. Insufficient evaluation of pain might contribute to these patients having less access to relevant healthcare services from which they could have benefited. While pain is a subjective experience, studies to objectively measure pain through physical metrics (e.g. cortisol levels, catecholamines, etc.) may aid in the assessment of pain. Although these metrics may be too expensive and time-consuming to be of use in the measurement of acute pain, they may serve to inform the development or validation of observational and self-report pain assessment tools. Given the heterogeneity of CP, future studies should seek to further classify patients based on the etiology, limb involvement, cognitive status, etc., as there may be differences evident in subsets of the population which may necessitate use of different treatment or assessment tools (Russo et al., 2008; Venkateswaran & Shevell, 2008). Efforts to characterize the experience of pain for patients with CP and, if necessary, to develop manageable objective pain assessment tools for this population - or to implement those that exist in clinically meaningful ways - are warranted.

Author statement

Nanfang Xu: Data curation; Formal analysis; Roles/Writing – original draft.

Hiroko Matsumoto: Investigation; Methodology; Project administration.

David Roye Jr.: Conceptualization; Funding acquisition; Writing – review & editing.

Joshua Hyman: Conceptualization; Supervision; Investigation.

Disclosures

The authors have stated that they had no interests which might be perceived as posing a conflict or bias.

Conflicts of interest and sources of funding

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

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