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ORIGINAL ARTICLE

Prevalence, identification, and interference of pain in young children with cerebral palsy: a population-based study

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ABSTRACT

Objective: To explore the presence of pain, how pain was addressed by physicians and parents, and how pain affected everyday life in young children with cerebral palsy (CP).

Methods: Children with CP, aged 5–10 years, participated in this cross-sectional study. Data were collected from medical records spanning a period of two years and by a standardized parental interview that included six structured questions and the Pain Interference Index.

Results: A total of 118 children, with a mean age of 7.4 years (SD 1.5), participated in the study. The parents of 81% of these children were interviewed. Pain was reported in 52% of the children, and pain was present at all severity levels. The prescription of analgesics was documented in 25% of these children's medical records. Fifty-nine percent of the children with pain received analgesics from their parents. Pain restricted the children's everyday lives particularly concerning sleep, school work and being with friends.

Conclusions: Half of this group of young children with CP were reported to have pain. Pain restricted the children's everyday lives and seemed to be under-treated. If pain can be addressed early, the children's everyday lives are likely to be improved.

➤ IMPLICATIONS FOR REHABILITATION

- There is a need to early identify and treat pain in young children.
- Important to discuss pain with parents irrespectively of the child's age and severity level.
- Pain interference assessment gives valuable information.
- Early treatment of pain might improve children's everyday life.

Introduction

Pain, the most common comorbidity in cerebral palsy (CP), has been identified as a central factor behind reduced quality of health and reduced participation in everyday activities reported by individuals with CP [1,2]. In a study exploring the health status of young adults with CP, pain, fatigue, and depression were present to a large extent, and pain interference was the only factor contributing to the variance in health status [3]. In children with CP, pain has been associated with social problems, school absence, reduced ambulation, more days spent in bed, low physical activity, and depression [4–9]. Moreover, pain affects not only the child and the family, but it contributes strongly to increases in societal health costs [10].
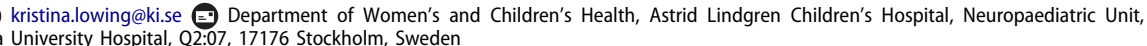
Pain prevalence has been reported to vary broadly, from 14% to 76%, in children and adults with CP [11]. Pain occurs in all CP subtypes and across all gross motor levels (GMFCS) [1,11–14]. More frequent pain has been reported in children with more extensive disability than in children with less severe CP [8,11,15,16]. Dystonia and hip subluxation/luxation were identified as the most common sources of pain, as was reported by

physicians in a cross-sectional study that included children and youth aged 3–19 years [12]. In children and young adults, musculoskeletal pain in the lower limbs has been reported to occur more frequently than pain in the upper limbs [11,17].

Although pain is common and is known to increase with age in children, young adults, and adults [11,18,19], pain seems to be both under-diagnosed and under-treated in CP [20,21].

Recent pain reviews have described difficulties in drawing conclusions about the presence of pain from existing studies, due to inclusion bias, heterogeneity in assessment tools and measurements, varying recall periods and varying age ranges of the participants. They recommended the inclusion of a well-defined age range of participants and the use of a standardized approach to pain assessment [11,22].

In a cross-sectional survey conducted in seven European countries, with the inclusion of 13- to 17-year-olds with CP ($n = 667$), the authors concluded that pain frequently occurred, and that strategies to reduce pain seemed absent or inadequate. The authors suggested a routine assessment of pain and the development of pain management plans [23]. Several authors have noted a need to identify and address pain early, so that the negative

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impact of pain on the quality of everyday life can be diminished or prevented [12,15,22,23].

Therefore, the overall aims of the present study were to explore the presence of pain in a population-based cohort of young children with CP, how the pain was identified and addressed by physicians and parents, and how the pain affected the everyday lives of these children.

More specifically, the study questions were:

1. What was the prevalence of pain in 5–10-year-old children with CP in a population-based Swedish cohort?
2. To what extent did the children with pain visit specialized physicians, and to what extent was the occurrence of pain recorded and addressed?
3. How did parents describe the frequency of pain in their children and the use of pain reducing drugs?
4. Did the pain interfere with the children's everyday lives?

Methods

Design

To address the study questions, an exploratory cross-sectional study design was used with a population-based inclusion of children with CP.

Study population and ethical approval

The study population consisted of children with CP, born from January 2007 to December 2011, and registered to live in the geographically defined central, north, and northwest areas of Stockholm. Prior to the study's beginning, an approval was granted by the Stockholm Regional Ethical Review Board (Dnr 2016/2424-31, 2017-01-26). Parents gave informed consent to participate.

Data collection

The data collection included two components: (1) for each child, medical records data from all visits with specialized pediatricians, child neurologists, and pediatric orthopedic surgeons (hereafter referred to as physicians) at the neuropediatric and pediatric orthopedic clinics at Karolinska University Hospital were gathered; and (2) a caregiver (parent) of the child was interviewed by telephone in a standardized manner.

Medical records

The investigator (MG) analyzed all notes from the outpatient visits during the preceding two years. Retrospective collection of two years' data was chosen to ensure inclusion of all children, including those with less frequent visits. The children's demographic data, subtype of CP, GMFCS level, cognitive level, and verbal communicative status were retrieved, as well as information on current seizure disorders. The verbal communicative status was classified in the following three levels: "no problem" implied that the child could understand and verbally communicate with familiar and unfamiliar persons, "some problem" implied that the child could understand and communicate some words with familiar persons, and "no verbal communication" implied that the child had no verbal communication. Emphasis was placed on identifying all mentions of pain in the medical record (i.e., whether pain had been discussed and documented at the visit). Additionally, the medical charts were explored to identify prescriptions for

analgesics. Only outpatient visits were included; data related to hospital admissions, visits as part of a surgical procedure, and any postoperative care were consequently not incorporated.

Telephone interviews

Letters with the study information were sent to the families. This included the option to decline the interview when contacted or to discontinue participation at any time during the interview. After a 1–5-week period, parents were contacted by telephone and given the opportunity to participate in the interview. All interviews were conducted from January 2017 to May 2017 by one investigator (MG).

Questionnaires

The interview included the Pain Interference Index (PII) and six study-specific structured questions, and the parents were asked to recall the situation during the previous two weeks. The PII contains six statements about how pain interferes with the child's everyday life (school, activities, friends, mood, mobility, and sleep). The response indicates how well the statement describes the child's present condition using a numerical scale, ranging from "not at all" (=0) to "very much" (=6) [24]. The questionnaire has been translated into Swedish, and its psychometric properties have been evaluated [25]. The six study-specific questions evaluated whether parents perceived that the child had experienced pain; how often the child had pain; the most common painful sites; if the parents had given the child any pain medication, and if so, how much; and last, whether the physician and parents had discussed pain during the prior two years when visiting the hospital.

Statistical analysis

Statistical analyses were performed with SPSS version 24 (SPSS Inc., Chicago, IL). Descriptive data are presented as frequency and percentage; median, interquartile range, and minimum and maximum are presented for ordinal or skewed continuous variables; and mean and standard deviation (SD) are presented for continuous variables. Parametric statistics (independent t-test) were used for interval and ratio data. Non-parametric statistics were used for ordinal data and data that did not demonstrate a normal distribution. In order to control for participant bias, the two independent-samples test was used; the Mann–Whitney U test was used to study the differences between the group reached by telephone and the group not reached. The chi-square test and the Mann–Whitney U test were used to study the differences between children who received and who did not receive prescriptions for analgesics. The significance level was set at $p < 0.05$. Spearman's correlation coefficient (r_s) was used to calculate correlations between the parent-reported pain frequency and how often the children with pain received analgesics from their parents, between the number of visits to the physicians and the number of prescriptions for analgesics, between how often the children with pain received analgesics from their parents and each of the following: CP subtype, GMFCS, level of cognition, communication disorder, presence of seizures, PII, sleep difficulties (PII), school work difficulties (PII), and difficulties in joining friends (PII).

Correlations were considered significant when $p < 0.05$ and $r_s > 0.30$. The following interpretation was used for the size of the correlation: r_s: 0.00–0.30 (negligible correlation), 0.30–0.50 (low

correlation), 0.50–0.70 (moderate correlation), 0.70–0.90 (high correlation), and 0.90–1.00 (very high correlation) [26].

Results

Study population characteristics

A total of 118 children with CP, 67 boys and 51 girls, lived in the catchment area. All children were included in the study, and the mean age was 7 years 4 months (SD: 1 year 4 months). All CP subtypes and GMFCS levels were represented (Table 1). Furthermore, the parents of 81% (95/118) of these children were reached by telephone and consented to participate in the telephone interview; the mean age of these children was 7 years 2 months (SD: 1 year 4 months) (Table 1). The mean age of the children (23/118) for which an interview was not conducted (not reached or declined) was 8 years 1 month (SD: 1 year 5 months) (mean difference: 11 months, 95% CI: 3–19, $p = 0.006$). Apart from the difference in age, the non-reached children did not differ significantly in any other aspect from those for which an interview was conducted (i.e., sex, CP type, GMFCS, communication level, cognition level, presence of seizures, total number of physician

Table 1. Descriptive statistics are presented for the total group of children, $n = 118$, and for the subgroup of children included in the parental telephone interview, $n = 95$, and for children not reached in the interview, $n = 23$.

	All children	Parental interview	Non-reached
Participants, n (%)	118	95 (81)	23 (19)
Sex, n (%)			
Male	67 (57)	56 (59)	11 (48)
Female	51 (43)	39 (41)	12 (52)
Age			
Mean (SD)	7.4 (1.5)	7.2 (1.4)	8.1 (1.5)
Min–max	5.1–9.9	5.1–9.9	5.1–9.8
GMFCS level, n (%)			
GMFCS I	62 (53)	49 (52)	13 (57)
GMFCS II	12 (10)	12 (13)	0
GMFCS III	12 (10)	10 (11)	2 (9)
GMFCS IV	9 (7.6)	5 (5)	4 (17)
GMFCS V	23 (19)	19 (20)	4 (17)
CP type, n (%)			
Unilateral	44 (37)	35 (37)	9 (39)
Bilateral	49 (42)	42 (44)	11 (48)
Dyskinetic	18 (15)	15 (16)	3 (13)
Ataxic	3 (2.5)	3 (3)	0
Seizures during the last 2 years, n (%)			
Yes	44 (37)	37 (39)	7 (30)
No	74 (63)	58 (61)	16 (70)
Cognition (IQ), n (%)			
>70	53 (45)	42 (44)	11 (48)
50–70	15 (13)	12 (13)	3 (13)
20–50	8 (7)	7 (7)	1 (4)
<20	23 (19)	20 (21)	3 (13)
No information[a]	19 (16)	14 (15)	5 (22)
Verbal communication, n (%)			
No problem	48 (41)	37 (39)	11 (48)
Some problem	39 (33)	33 (35)	6 (26)
Non verbal	31 (26)	25 (26)	6 (26)
Visits to physician	483	398	85
Median (25th–75th)	4 (2–5)	4 (2–5)	4 (3–5)
Min–max	0–11	0–11	0–9
Visits with a documented pain-discussion			
Median (25th–75th)	0 (0–1)	0 (0–1)	0 (0–1)
Min–max		0–6	0–6	0–4
Visits with analgesics prescribed, n (%)			
Yes	23 (19)	17 (18)	6 (26)
No	95 (81)	78 (82)	17 (74)

Data are presented as number (n), percentage (%), mean, standard deviation (SD), median and quartiles (25th–75th), and minimum–maximum (min–max).
[a]Not formally tested and have not yet started school.

visits, visits with pain discussion, or prescription of analgesics) (Table 1).

Prevalence of pain registered in the medical records

Data retrieved from the hospital records of the 118 children during the two-year time period displayed a total of 483 visits to physicians. Pain (present or absent) was documented in the medical record in 19% ($n = 92/483$) of these visits, belonging to 43% ($n = 51/118$) of the children.

During the two-year period, 19% of the children ($n = 23/118$) were given or prescribed drugs by their physician specifically to alleviate pain. NSAIDs and paracetamol were used, but opioids were never used. Most often, the pain was considered to be muscle tone related, and diazepam, clonidine or botulinum toxin-A was also prescribed or administered as analgesics. A higher frequency of visits to the physicians was observed in the group of children ($n = 23/118$) who received prescriptions for analgesics (mean: 6.2 [SD 2.2]) compared to the group of children ($n = 95/118$) without prescriptions (mean: 3.6 [SD 2.4]) (mean difference: 2.6, 95% CI: 1.5–3.7, $p < 0.001$). Concerning prescriptions for analgesics, significant differences were observed between the CP subtypes: 10/18 children with dyskinetic CP, 10/53 with bilateral CP, 3/44 with unilateral CP, and none with ataxic CP received prescriptions (chi-square: 19.9, df: 3, $n = 118$, $p < 0.001$). Differences were also observed between GMFCS levels: 14/23 children in GMFCS V, 2/9 in GMFCS IV, 2/12 in GMFCS III, 2/12 in GMFCS II, and 3/62 in GMFCS I received prescriptions (chi-square: 33.5, df: 4, $n = 118$, $p < 0.001$). Among the 23/118 children who received prescriptions for analgesics, seizures were present in 15/23 children, intellectual disability was present in 14/23 children and communication disorder was present in 19/23 children. Sex (boys = 15/23 and girls = 8/23) (chi-square: 0.822, df: 1, $n = 118$, $p = 0.365$) and age (mean difference: 0.4, 95% CI: –0.3 to 1.1, $p = 0.289$) did not differ with respect to prescriptions for analgesics.

Telephone interview: parent-reported pain during the previous two-week period

Parents of 52% of the children ($n = 49/95$) reported pain during the previous two-week period; daily pain was reported in 37%, pain several times a week in 33% and pain once a week in 30% of these children. Parents described the most common location of pain as the lower limbs (71%). Children with pain during the previous two-week period had a higher frequency of visits to a physician (during the two-year period) in comparison to the group of children without pain (pain: mean 4.8 [SD 2.6], no pain: mean 3.5 [SD 2.6], mean difference: 1.3, 95% CI: 0.27–2.4, $p = 0.014$). In 53% of the children with pain ($n = 26/49$), documentation of pain in the medical records was observed. Parents of 51% of the children with pain ($n = 25/49$) reported that pain had been addressed at visits to the physicians during the two-year period. A prescription for analgesics was documented in 25% ($n = 12/49$) of these children's medical records. During the previous two weeks, analgesics (prescription and/or over-the-counter) had been given by the parents to 59% of the children with pain. More specifically, 4% received physician-prescribed analgesics, 16% received both physician-prescribed and over-the-counter analgesics and 39% received over-the-counter analgesics, while 41% of the children had not received any pain reliever from their parents. No significant differences were observed between the children with and without parent-reported pain in relation to sex, age, CP subtype, GMFCS, presence of seizures, level of cognition, communicative disorder, or prescriptions for analgesics (Table 2).

Table 2. Descriptive data from the children included in the parental telephone interview ($n = 95$), with 49 children with pain during the last two-week period and 46 children without pain.

$n = 95$	Children with pain, $n = 49$	Children without pain, $n = 46$	Children with/without pain
Age, mean (SD)	7.29 (1.5)	7.08 (1.4)	($z=-0.749$) $p = 0.454$
Sex, n (%)			($z=-0.048$) $p = 0.962$
Boy	29 (59)	27 (59)	
Girl	20 (41)	19 (41)	
CP type, n (%)			($z=-0.141$) $p = 0.888$
Unilateral	18 (37)	17 (37)	
Bilateral	17 (35)	21 (46)	
Dyskinetic	13 (27)	6 (13)	
Ataxic	1 (2)	2 (4)	
GMFCS, n (%)			($z=-1.509$) $p = 0.131$
I	23 (47)	26 (56)	
II	7 (14)	5 (11)	
III	3 (6)	7 (15)	
IV	0	5 (11)	
V	16 (33)	3 (7)	
Seizures during the last two years, n (%)			($z=-1.640$) $p = 0.101$
Yes	23 (47)	14 (30)	
No	26 (53)	32 (70)	
Cognition (IQ), n (%)			($z=-0.615$) $p = 0.539$
>70	23 (47)	19 (41)	
50–70	4 (8)	8 (17)	
20–50	3 (6)	4 (9)	
<20	14 (29)	6 (13)	
No information[a]	5 (10)	9 (20)	
Verbal communication, n (%)			($z=-0.643$) $p = 0.520$
No problem	21 (43)	16 (35)	
Some problem	10 (20)	23 (50)	
Non verbal	18 (37)	7 (15)	
Visits to physicians			($z=-2.579$) $p = 0.010$
Median (25th–75th)	4 (3–6)	3 (1.75–5)	
Min–max	1–11	0–11	
Visits where pain discussion documented			($z=-2.018$) $p = 0.044$
Median (25th–75th)	0 (1–1)	0 (0–1)	
Min–max	0–4	0–6	
Medical pain treatment, n (%)			($z=-1.722$) $p = 0.085$
Yes	12 (25)	5 (11)	
No	37 (75)	41 (89)	

Data are presented as mean and standard deviation (SD), median and quartiles (25th–75th), number (n), and percentage (%). Differences between children with pain and children without pain are calculated with the Mann–Whitney test and presented with (z-value) p values.
[a]Not formally tested and have not yet started school.

Pain interference in everyday life

Pain interference during the previous two-week period was assessed by the PII. It demonstrated a higher interference in the group of children with pain (mean: 8.7 [SD 8.9], $n = 49$) in comparison to the children without pain (mean: 1.7 [SD 4.9], $n = 46$) (mean difference: 7, 95% CI: 4–9.9, $p < 0.001$). Pain affected the children's everyday lives, with sleep, school work, and problems being with friends reported in particular.

Associations between the presence of pain, pain treatment, physician visits, and comorbidities in children with pain reported by parents (n = 49)

Among the children with pain during the previous two-week period, an expected association was found between how often the children had pain and how often the children received analgesics from their parents ($r_s = 0.48$, $p < 0.001$). An association was also present between the number of visits to the physicians and prescriptions for analgesics ($r_s = 0.37$, $p = 0.008$). How often the parents gave their children analgesics was associated with the children's presence of comorbidities and the pain interference in the children's everyday lives (Table 3).

Table 3. Spearman's correlation coefficient (r_s) was calculated for associations between how often the children with pain received analgesics from their parents and the following factors ($n = 43$–48).

$n = 43$–48	How often children received analgesics	p
CP type	$r_s=0.45$ ($n = 48$)	$p = 0.002$
GMFCS	$r_s=0.50$ ($n = 48$)	$p < 0.001$
Level of cognition	$r_s=0.54$ ($n = 43$)	$p < 0.001$
Communication disorder	$r_s=0.51$ ($n = 48$)	$p < 0.001$
Presence of seizures	$r_s=0.60$ ($n = 47$)	$p < 0.001$
Pain interference index (PII)	$r_s=0.56$ ($n = 48$)	$p < 0.001$
Sleep difficulties (PII)	$r_s=0.61$ ($n = 49$)	$p < 0.001$
School-work difficulties (PII)	$r_s=0.56$ ($n = 48$)	$p < 0.001$
Difficult to join friends (PII)	$r_s=0.51$ ($n = 48$)	$p < 0.001$

Discussion

The major findings of this study were that pain, present in more than half of the 5–10-year-old children with CP, occurs early and at all severity levels. Children with pain had a higher frequency of visits to the physician, but documentation in the medical records and prescription of analgesics occurred less often. Interference of pain with the children's everyday lives was evident.

The present study is the first to report the presence and interference of pain in a population of children aged 5–10 years with CP. The age of interest was chosen due to several factors.

First, pain has been known to increase with age, but many previous studies included a wide age range of children, thus obscuring how common the problem is in the younger child. Second, parents of young children typically still have close contact and a good knowledge of the child's current situation. The older and more independent child might not inform parents of possible problems to the same extent. Third, from a clinical viewpoint, we also believe that knowledge of the expected prevalence of pain in the younger child can motivate physicians and parents to be more observant of the signs of pain and to consider treatment. Early identification and treatment will then reduce the likelihood of the child developing a chronic, more difficult to treat, pain condition.

In addition, the inclusion of a well-defined age range of participants and the use of a standardized approach to pain assessment have recently been recommended [11,22].

Higher pain frequency has been reported in children with more extensive disability than in children with less severe CP [8,11,15,16]. In the present study, the physicians to a greater extent prescribed analgesics to children who were more severely affected (dyskinetic CP, GMFCS V, presence of seizures, intellectual, and communicative disabilities). In contrast, when the parents reported their children's pain, it was common at all severity levels. As an example, pain was reported in 50% of the children with unilateral CP and almost 40% in children at GMFCS I. However, moderate associations were detected between how often children received analgesics from their parents and the children's extent of comorbidities and pain interference in everyday life. Overall, these results indicate that the less severely affected children also have pain, but to a lesser extent receive analgesics, both concerning the prescription of analgesics and when given by their parents.

In the present study, age and sex did not influence the presence of pain reported by parents or pain documented in the medical records. Conflicting results have earlier been reported as to whether age and sex affect the occurrence of pain in children and adolescents with CP [17,19,27]. Riquelme et al. did not observe any influence of age in children, youth, and young adults (6–30 years) with CP, while the influence was evident in their group of healthy controls with pain [27]. In contrast, the authors of a retrospective study including children aged 1–14 years observed pain more often in girls, and their pain frequency increased with age [19]. Ramstad et al. explored the presence of pain in children and adolescents (aged 8–18 years) with CP, and they found that age above 14 years was the only significant predictor [17]. According to the evidence presented above, the age range (5–10 years) in our population might have been too narrow and too low to detect whether age and sex affect pain, and it may explain why we did not observe such an influence.

The most common pain location in our young population was the lower extremities, reported in more than 70% of the children. During the interview, the parents were not asked to give their opinions as to the most likely cause of the pain. However, from the medical records, it was clear that muscle tone was perceived as a common etiology for pain. Other studies have similarly reported that pain was located more frequently in the lower limbs and less often in the upper limbs [11,12,19]. These studies have suggested that the etiology of pain is multifaceted and includes factors such as abnormal biomechanical forces, high muscle tone, muscle overuse, and hip subluxation/luxation [11,12,19].

In a recent review, the authors recommended the use of validated measures that assess not only the intensity and location of pain, but also the possible interference of pain in the children's everyday lives [28]. In our study, pain interference was explored, and parents reported that pain clearly restricted the children's daily lives. Pain interference was particularly marked concerning sleep disturbances, problems with school work and in difficulties being with friends. For older children, findings consistent with this have earlier been reported, such as absence from school, lower school functioning (due to pain-related fatigue), reduced ambulation, higher frequencies of days spent in bed and less participation [4,5,7,8,29]. Pain intensity has been associated with emotional and behavioral problems, whereas the anxiety or fear related to experiencing pain has been less explored but was reported to be the predictor that made a significant and independent contribution to depressive disorder in children with CP aged 5–15 years [4].

Pain has indeed been identified as a major contributor to decreased participation in everyday activities and reduced quality of life in CP [1,2]. In a recent cross-sectional study including a cohort of youth with CP, where factors associated with pain were explored, the authors concluded that if a youth's everyday activities deteriorated, one should primarily consider pain as the cause of the decline before planning other interventions [1]. Positive effects on children's pain severity were reported when initially caregiver-rated pain was communicated with the physician and treated [30]. These are facts highlighting the importance of thoroughly discussing pain issues with parents and children.

Apart from affecting the child and his or her family, pain remains a significant societal economic burden. In an American study aiming to assess the impact of pediatric pain-related conditions on health care expenditures, the authors discovered that the costs related to pain in children exceeded the costs for children with asthma and obesity. They concluded that efforts to prevent and treat pediatric pain-related conditions are immediately needed [10].

Strengths and limitations

Strengths of the present study include the use of a total population of children aged 5–10 years and a standardized approach with a predesigned protocol that was applied both in the exploration of the medical records and in the telephone interviews. For pain interference, a validated questionnaire was used. Another strength is the high number of participants in the telephone interview, resulting in a truly population-based cohort and reducing the risk of recruitment bias. In addition, the included population appears to be representative in relation to sex and distribution of CP subtypes and GMFCS levels, even though the proportion of children classified in GMFCS I was slightly higher than the proportion reported in the 2018 Swedish National Cerebral Palsy Registry [19,31,32]. The use of parent-reported pain questions could be regarded as a limitation since self-reported pain has been considered the gold standard. However, younger children may be unable to provide reliable self-reports due to limitations by developmental stages, cognitive disability or communicative disorder [33]. Therefore, to use parental reports of pain could sometimes be considered the most appropriate approach [34]. A limitation is that the cognitive level had not yet been formally tested by a psychologist in all children, possibly underestimating the true amount of intellectual disability. In addition, the study aimed to identify non-procedural pain since the availability of surgery or injections with botulinum toxin-A, for example, can vary in relation to external factors. One can argue that to exclude procedural pain underestimates the true pain experienced in some children with CP.

In conclusion, pain was present in more than half of this young population and occurred at all severity levels. There was an under-treatment of pain, especially in less severely affected children. Prescriptions for analgesics, and administration of analgesics by the parents, occurred more frequently in severely affected children. Pain restricted the children's everyday lives and was especially limiting concerning sleep, school work, and the ability to spend time with friends. It is a major and important challenge to identify and address pain early enough to prevent or mitigate the negative impact of pain on the everyday life of each child.

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Disclosure statement

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References

[1] Fairhurst C, Shortland A, Chandler S, et al. Factors associated with pain in adolescents with bilateral cerebral palsy. Dev Med Child Neurol. 2018;61(8):929–936.

[2] Findlay B, Switzer L, Narayanan U, et al. Investigating the impact of pain, age, Gross Motor Function Classification System, and sex on health-related quality of life in children with cerebral palsy. Dev Med Child Neurol. 2016;58(3):292–297.

[3] Sienko SE. An exploratory study investigating the multidimensional factors impacting the health and well-being of young adults with cerebral palsy. Disabil Rehabil. 2018;40(6):660–666.

[4] Yamaguchi R, Nicholson Perry K, Hines M. Pain, pain anxiety and emotional and behavioural problems in children with cerebral palsy. Disabil Rehabil. 2014;36(2):125–130.

[5] Russo RN, Miller MD, Haan E, et al. Pain characteristics and their association with quality of life and self-concept in children with hemiplegic cerebral palsy identified from a population register. Clin J Pain. 2008;24(4):335–342.

[6] Tervo RC, Symons F, Stout J, et al. Parental report of pain and associated limitations in ambulatory children with cerebral palsy. Arch Phys Med Rehabil. 2006;87(7):928–934.

[7] Berrin SJ, Malcarne VL, Varni JW, et al. Pain, fatigue, and school functioning in children with cerebral palsy: a path-analytic model. J Pediatr Psychol. 2006;32(3):330–337.

[8] Houlihan CM, O'Donnell M, Conaway M, et al. Bodily pain and health-related quality of life in children with cerebral palsy. Dev Med Child Neurol. 2004;46(5):305–310.

[9] Whitney DG, Warschausky SA, Peterson MD. Mental health disorders and physical risk factors in children with cerebral palsy: a cross-sectional study. Dev Med Child Neurol. 2019;61(5):579–585.

[10] Groenewald CB, Wright DR, Palermo TM. Health care expenditures associated with paediatric pain-related conditions in the United States. Pain. 2015;156(5):951–957.

[11] McKinnon CT, Meehan EM, Harvey AR, et al. Prevalence and characteristics of pain in children and young adults with cerebral palsy: a systematic review. Dev Med Child Neurol. 2019;61(3):305–314.

[12] Penner M, Xie WY, Binepal N, et al. Characteristics of pain in children and youth with cerebral palsy. Pediatrics. 2013;132(2):e407–e413.

[13] Novak I, McIntyre S, Morgan C, et al. A systematic review of interventions for children with cerebral palsy: state of the evidence. Dev Med Child Neurol. 2013;55(10):885–910.

[14] Palisano R, Rosenbaum P, Walter S, et al. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Dev Med Child Neurol. 1997;39(4):214–223.

[15] Westbom L, Rimstedt A, Nordmark E. Assessments of pain in children and adolescents with cerebral palsy: a retrospective population-based registry study. Dev Med Child Neurol. 2017;59(8):858–863.

[16] Barney CC, Krach LE, Rivard PF, et al. Motor function predicts parent-reported musculoskeletal pain in children with cerebral palsy. Pain Res Manag. 2013;18(6):323–327.

[17] Ramstad K, Jahnsen R, Skjeldal OH, et al. Characteristics of recurrent musculoskeletal pain in children with cerebral palsy aged 8 to 18 years. Dev Med Child Neurol. 2011;53(11):1013–1018.

[18] Benner JL, Hilberink SR, Veenis T, et al. Long-term deterioration of perceived health and functioning in adults with cerebral palsy. Arch Phys Med Rehabil. 2017;98(11):2196–2205.e1.

[19] Alriksson-Schmidt A, Hagglund G. Pain in children and adolescents with cerebral palsy: a population-based registry study. Acta Paediatr. 2016;105(6):665–670.

[20] Baxter P. Comorbidities of cerebral palsy need more emphasis-especially pain. Dev Med Child Neurol. 2013;55(5):396.

[21] Fehlings D. Pain in cerebral palsy: a neglected comorbidity. Dev Med Child Neurol. 2017;59(8):782–783.

[22] Ostojic K, Paget SP, Morrow AM. Management of pain in children and adolescents with cerebral palsy: a systematic review. Dev Med Child Neurol. 2019;61(3):315–321.

[23] Parkinson KN, Dickinson HO, Arnaud C, et al. Pain in young people aged 13 to 17 years with cerebral palsy: cross-sectional, multicentre European study. Arch Dis Child. 2013;98(6):434–440.

[24] Martin S, Nelson Schmitt S, Wolters PL, et al. Development and validation of the English Pain Interference Index and Pain Interference Index-Parent report. Pain Med. 2015;16(2):367–373.

[25] Holmstrom L, Kemani MK, Kanstrup M, et al. Evaluating the statistical properties of the pain interference index in children and adolescents with chronic pain. J Dev Behav Paediatr. 2015;36:450–454.

[26] Hinkle DE, Wiersma W, Jurs SG. Applied statistics for the behavioral sciences. Boston (MA): Houghton Mifflin; University of Michigan; 2003.

[27] Riquelme I, Cifre I, Montoya P. Age-related changes of pain experience in cerebral palsy and healthy individuals. Pain Med. 2011;12(4):535–545.

[28] Schiariti V, Oberlander TF. Evaluating pain in cerebral palsy: comparing assessment tools using the International

Classification of Functioning, Disability and Health. Disabil Rehabil. 2018;1–8.

[29] Novacheck TF, Stout JL, Tervo R. Reliability and validity of the Gillette Functional Assessment Questionnaire as an outcome measure in children with walking disabilities. J Paediatr Orthop. 2000;20(1):75–81.

[30] Christensen R, MacIntosh A, Switzer L, et al. Change in pain status in children with cerebral palsy. Dev Med Child Neurol. 2017;59(4):374–379.

[31] Himmelmann K, Uvebrant P. The panorama of cerebral palsy in Sweden part XII shows that patterns changed in the birth years 2007–2010. Acta Paediatr. 2018;107(3): 462–468.

[32] CPUP. 2018. Available from: http://cpup.se/wp-content/uploads/2018/10/%C3%85rsrapport-CPUP-20181025.pdf

[33] Franck LS, Greenberg CS, Stevens B. Pain assessment in infants and children. Pediatr Clin North Am. 2000;47(3): 487–512.

[34] Houlihan CM, Hanson A, Quinlan N, et al. Intensity, perception, and descriptive characteristics of chronic pain in children with cerebral palsy. J Pediatr Rehabil Med. 2008;1(2): 145–153.