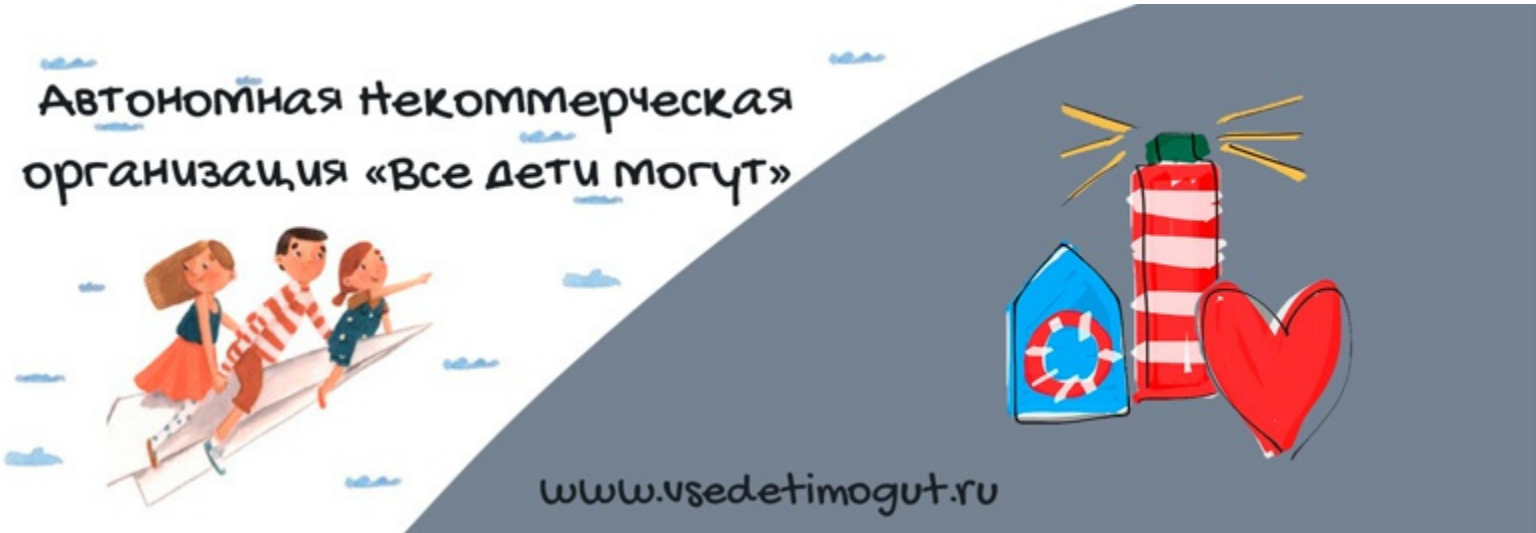
Боль у детей и подростков с церебральным параличом



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REGULAR ARTICLE

Pain in children and adolescents with cerebral palsy: a population-based registry study

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ABSTRACT

Aim: We assessed prevalence and location of pain in a total population of children and adolescents with cerebral palsy (CP) based on the Gross Motor Function Classification System (GMFCS), age and gender.

Methods: This cross-sectional study was based on the last assessment of children aged 1–14 years in the combined Swedish follow-up programme and national quality register programme for CP. All were born 2001–2012 and reported to the registry in 2013–2014. Logistic regression was used to regress age, gender and the GMFCS level on the presence of pain. We also assessed pain sites among GMFCS groups.

Results: We included 2777 children (57% boys) at a median age of 7 years; 32.4% reported pain, with significantly more girls than boys experiencing pain and significantly more children at GMFCS levels III and V than GMFCS I. Pain frequency increased with age and differences among GMFCS levels were found in the lower extremities and abdomen. Pain in the abdomen and hips was most frequent at GMFCS V, knee pain at level III and foot pain at level I.

Conclusion: Our results showed that although a lower prevalence than in many other studies, pain constituted a significant problem in children and adolescents with CP.

INTRODUCTION

Cerebral palsy (CP) is associated with progressive musculoskeletal complications and reduced participation in society (1,2). Pain is a secondary complication of CP and is receiving more attention, although research on the aetiologies and pathways that lead to pain in individuals with CP, and on effective treatment approaches, remain scarce (3–8). Norwegian researchers have reported that recurrent musculoskeletal pain is common and associated with higher levels of mental health problems and worse health-related quality of life (5), a finding that has been replicated in Canada (7). In the Study of Participation of Children with Cerebral Palsy Living in Europe (SPARCLE), a European multinational longitudinal study on CP, 54% of children with CP reported having experienced pain in the previous week and this was also associated with poorer quality of life (4). Pain was even more frequent in a subsequent SPARCLE follow-up study in adolescence, where 75% of the participants reported that they experienced pain in a typical week and with girls reporting pain more often than boys (6). According to a systematic review on comorbidities, diseases and functional limitations associated with CP, 75% were in pain (9). The need for population-based studies to investigate the presence of pain in individuals with CP was particularly stressed (9), and the need for more research on pain in CP in general has also been highlighted (10).

Levels of functioning and comorbidities vary greatly in individuals with CP. In the last few decades, it has become increasingly common to classify the gross motor function of individuals with CP according to the Gross Motor Function Classification System (GMFCS). The GMFCS is a psychometrically sound instrument used by both clinicians and researchers to classify gross motor function based on self-initiated movement. There are five levels, with level one

Abbreviations

CP, Cerebral palsy; CPUP, Cerebral Palsy Follow-Up Program; GMFCS, Gross Motor Function Classification System; SCPE, Surveillance of Cerebral Palsy Network in Europe; SPARCLE, Study of Participation of Children with Cerebral Palsy Living in Europe.

Key notes

- We assessed the prevalence and location of pain in 2777 children and adolescents with cerebral palsy (CP), using Swedish registry data and focusing on motor function, age and gender.
- Just under a third (32.4%) of the children reported pain, and the frequency was higher in girls and older children.
- Pain was frequently reported at all Gross Motor Function Classification System levels, but the pain site profiles differed.

describing the highest level of function and level five the lowest (11,12).

For more than 20 years, children and adolescents living with CP in Sweden have been eligible to participate in a combined follow-up programme and national quality register called the *Cerebral Palsy Follow-Up Program* (CPUP). CPUP originated in 1994 in the south of Sweden, but the entire nation has been included since 2005, when the programme also became a national quality register. CPUP is a multidisciplinary secondary prevention programme used by the Swedish habilitation services to detect early signs of deterioration. Specialties include orthopaedic surgeons, hand surgeons, physiotherapists, occupational therapists and neuropaediatricians. Social workers, speech therapists, psychologists, special educators and dieticians are part of the habilitation teams if required. Coverage rates are high, and virtually, the entire population of children and adolescents with CP in Sweden is included. Data on adults with CP are available but to date these are not population based as adults were not included in CPUP until 2011. The programme adheres to the aspirations of the World Health Organization's *International Classification of Functioning Disability and Health* (13), a biopsychosocial model that emphasises activities and participation.

In this study, we investigated the presence of pain, the site or sites of pain and how these related to gender, gross motor function and age. It was hypothesised that pain would be more frequent in females and at older ages. We also hypothesised that the site of the pain profile would differ based on the gross motor function level.

PARTICIPANTS AND METHODS

This was a cross-sectional study based on CPUP data from the most recent visit of all children born between 2000 and 2012 who were reported to the registry in 2013–2014. The CP diagnosis was determined by a neuropaediatrician according to the Surveillance of Cerebral Palsy Network in Europe (14). In CPUP, children at GMFCS level I are examined by their physiotherapist annually up to 6 years of age and then every second year. Those at GMFCS levels II–V are examined twice a year up to 6 years, then once a year. In addition to a physical assessment, the physiotherapist completes a general survey that asks whether the child or their parents have stated that the child is in pain. If the answer is yes, a follow-up question is asked about where it hurts. If the child is able to communicate, he or she will answer, if not the parent or legal guardian answers the question.

Age was calculated based on the date of birth and last visit date and was recorded as a continuous variable in years. Gender was recorded as a dichotomous variable, namely male or female. The extended and revised version of the GMFCS (12) was used to classify *gross motor function* and the physiotherapist categorised the participant as GMFCS level I–V. Pain was dichotomised as present or not present. The site or sites of pain were recorded as head, neck, back, arms, hands, hips, knee, feet, teeth, stomach, pressure, skin wound or other. For the purposes of our analyses we reclassified these categories by combining the head and neck, the arms and hands, the thighs and hips and the lower legs and feet. Missing data on the site of the pain were coded as no in the analyses. We also classified whether the participant experienced pain in one or multiple sites.

Statistical analysis

Raw numbers and percentages were calculated on all variables. Logistic regression was used to regress age, gender and the GMFCS level on the presence of pain. An adjusted logistic regression on the GMFCS level and presence of pain, adjusted for age and gender, was also performed. We used 95% confidence intervals (95% CIs) to assess statistical significance among GMFCS groups on pain sites.

The study was approved by the Ethics Board at Lund University.

RESULTS

A total of 2777 children with a median age of 7 years, standard deviation (SD) of 3.6 years and range of one to 14 years were included in the study. The gender and corresponding GMFCS distribution are presented in Table 1.

The child was asked, or their parent or guardian if they could not communicate, if the child experienced pain and 900 (32.4%) said yes. A further 1799 (64.8%) said no and 78 (2.8%) had missing data on this specific item. Males (30.1%) were significantly less likely to report pain than females (35.5%) (OR = 0.79, 95% CI 0.67–0.94). The proportion of children with pain increased with age, from 17% of children of 2 years of age to 50% of children of 14 years of age (OR = 1.12, 95% CI 1.09–1.14) (Fig. 1). After adjusting for age and gender, children at GMFCS levels III (OR = 1.40, 95% CI 1.02–1.84) and V (OR = 1.52; 95% CI 1.20–1.92) were significantly more likely to report pain than those at GMFCS level I.

Data on the site or sites of pain were available for 829 of the 900 children (92.1%) who experienced pain and 175 children (19.4%) experienced pain at multiple sites: 5.8% of the total population at GMFCS I, 6.3% at GMFCS II, 9.3% at GMFCS III, 6.3% at GMFCS IV and 5.9% at GMFCS V. No statistically significant differences were found based on the GMFCS level or gender.

Pain was most common in the lower extremities (n = 624). Of the 829 who reported pain sites, 325 (36.1%) reported pain in the feet, 193 (21.4%) reported knee pain, 263 (29.2%) reported pain in the hips, 97 (10.8%) had pain in the abdomen, 84 (9.3%) reported back pain, 83 (9.2%) in the head/neck, and 81 (9%) had pain in the arms/hands. Fewer than 15 children reported pain from teeth, pressure, skin wounds or other sites. Statistically significant differences based on GMFCS level were found on pain in the lower extremities, namely the feet, knee and hips (Fig. 2) and abdomen (Fig. 3). Pain in the abdomen

Table 1 Distribution of age, gender and gross motor function classification system in population of children and adolescents with cerebral palsy

		Gender		GMFCS level				
Age (years)	Number of children	Boys (%)	Girls (%)	I (%)	II (%)	III (%)	IV (%)	V (%)
1–2	297	166 (56)	131 (44)	95 (32)	63 (21)	37 (12)	58 (20)	44 (15)
3–4	469	263 (56)	206 (44)	228 (49)	68 (14)	55 (12)	50 (11)	68 (14)
5–6	492	298 (61)	194 (39)	224 (46)	75 (15)	41 (8)	73 (15)	79 (16)
7–8	454	267 (59)	187 (41)	190 (42)	75 (17)	47 (10)	69 (15)	73 (16)
9–10	429	250 (58)	179 (42)	203 (47)	68 (16)	25 (6)	69 (16)	64 (15)
11–12	424	232 (55)	192 (45)	167 (39)	75 (18)	38 (9)	58 (14)	86 (20)
13–14	212	119 (56)	93 (44)	93 (44)	39 (18)	15 (7)	38 (18)	27 (13)
Total	2777	1595 (57)	1182 (43)	1200 (43)	463 (17)	258 (9)	415 (15)	441 (16)

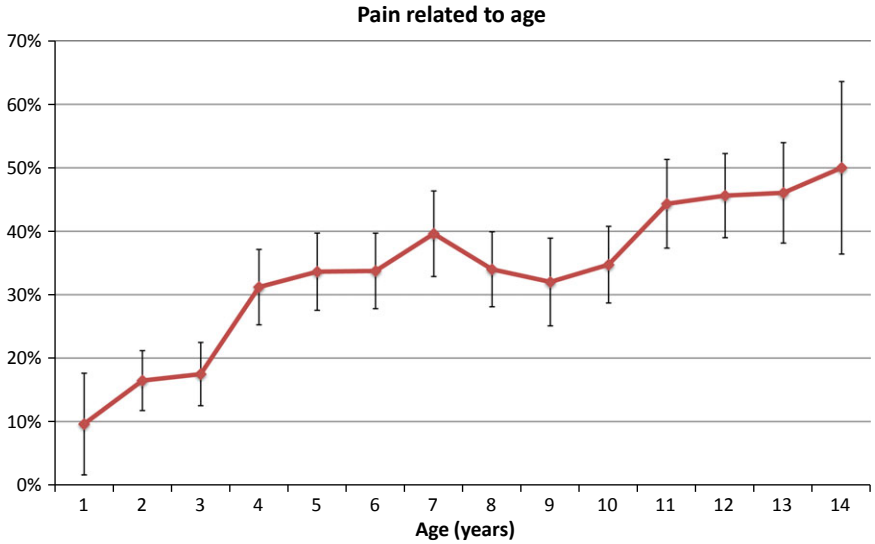

Figure 1 Proportion of children and adolescents with cerebral palsy reporting pain based on age. (The line segments represent the upper and lower bounds of the 95% confidence interval).

and, or, hips was most frequent at GMFCS level V, knee pain at level III and pain in the feet at level I. No statistically significant differences were found based on the GMFCS level on arm, head or neck or back pain.

DISCUSSION

About one-third of the children and adolescents with CP in Sweden reported pain at their last CPUP assessment. Although this level was high, it was still lower than the 50-75% reported by other CP studies (4,6,8,9,15). A number of explanations can account for this apparent discrepancy. The item in CPUP used to determine the presence of pain is a broad screening item that does not specify duration, frequency or severity. Pain instruments used in other studies may have been more specific and assessed pain during a specific time period. This may have resulted in a higher prevalence than the level found in our study. Some of the other studies focused on specific types of pain, such as musculoskeletal pain (8), or included specific subtypes of CP, for example bilateral spastic (16), both of which could influence the prevalence rate. Furthermore, we included a wide age span, one to 14 years of age, whereas the other studies focused more on pain in adolescents with CP (6). Our results did indicate that pain prevalence increased with age.

Three regions of Sweden did not include children born in 2000–2001 in CPUP, explaining the lower number of 13 and 14 year olds in our study than would be expected. Our findings are based on data from the total population of children and young adolescents, reducing the likelihood of a selection effect. The GMFCS distribution, with 43% of children in GMFCS level I, agreed with previous epidemiologic studies (17). Most of the other studies had included a lower proportion of children in GMFCS I. To our knowledge, this study is unique in that it represents the actual GMFCS distribution found in the population of children and young adolescents with CP. It is difficult to achieve a

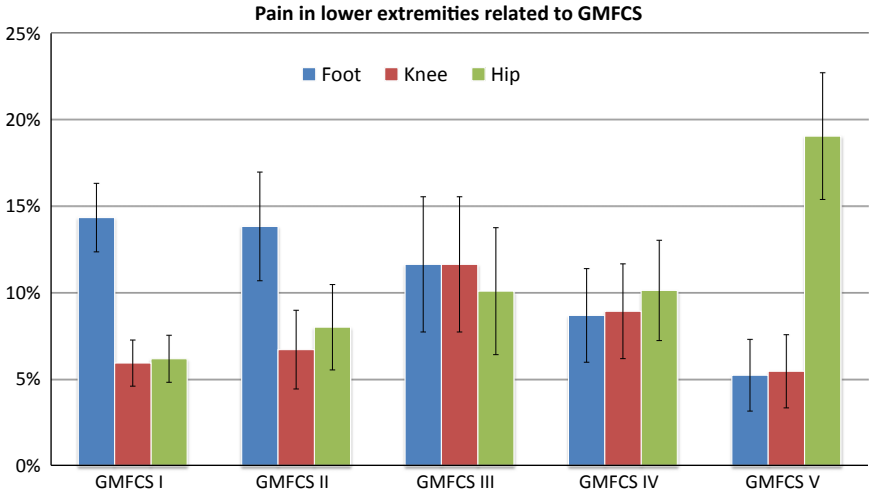

Figure 2 Proportion of pain reported in the lower extremities – feet and or lower leg, knee, hips and or thigh – in children and adolescents with cerebral palsy based on the gross motor function classification system levels. The line segments represent the upper and lower bounds of the 95% confidence interval.

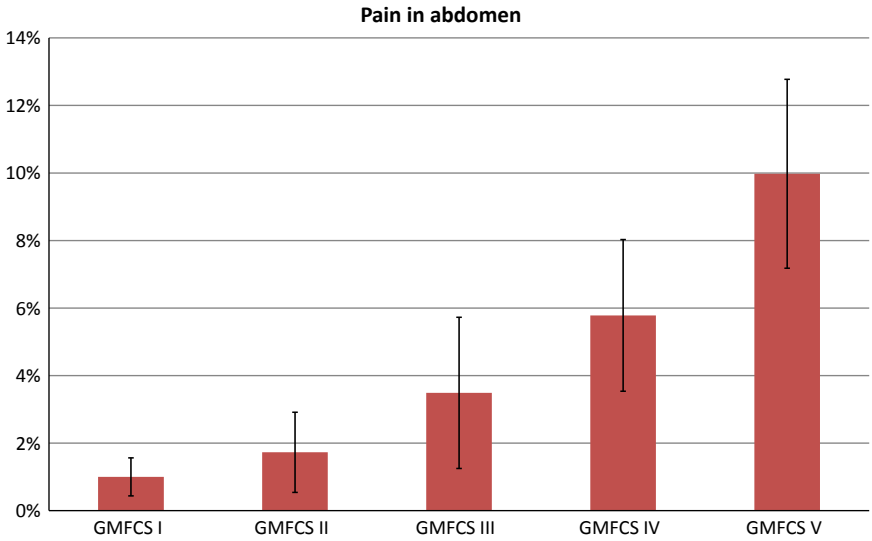

Figure 3 Proportion of pain reported in the abdomen in children and adolescents with cerebral palsy based on the gross motor function classification system. The line segments represent the upper and lower bounds of the 95% confidence interval.

representative sample in pain studies, and it is plausible that individuals who are actually in pain may be more interested in participating. CPUP includes virtually all children and adolescents with CP in Sweden and does not specifically target pain. Consequently, it is possible that we were also more likely to capture those who did not experience pain. Those in GMFCS III and GMFCS V were more likely to report pain than those in GMFCS I. This makes intuitive sense, yet previous results have been inconsistent. Dickinson et al. (4) and Rethlefsen et al. (18) found no significant association between the GMFCS level and the presence of pain, whereas Penner et al. (3), who used reports by parents and physicians, did find significant associations.

In terms of gender differences, pain was more commonly reported in girls than boys and prevalence increased with age. Gender differences in this direction have been noted consistently (6,19). For example, the likelihood of pain in the knees has been reported to be five times higher in females than males with CP (18). Although chronic pain did not differ between males and females with spastic bilateral CP, chronic pain that lasted more than 1 year was significantly more common in females in a Dutch study (16). Increased rates of pain at older ages have also been reported previously (3,19).

With regard to the site of pain, it occurred most frequently in the lower extremities and we also found

associations between the pain site and the GMFCS levels. Individuals in GMFCS I and II were more likely to report pain in the feet or lower legs than those in GMFCS level V. Those in GMFCS I-II were ambulant, as opposed to those in GMFCS V, and therefore, they had the greatest load on the feet and lower leg muscles. Pain in the knees was more frequent in those in GMFCS III than GMFCS I and V and this could be because crouch gait is most frequent in GMFCS III (20). The abnormal biomechanical forces in crouch gait might result in pain due to excessive stress and overuse (20). Individuals at GMFCS V were more likely to have pain in the hips and thighs than any other GMFCS level. Hip dislocation is a common cause of hip pain in CP (21,22), but there were only 13 children with hip dislocations in the data as a consequence of CPUP hip surveillance programme. Since the initiation of the CPUP programme, hip dislocations in Sweden have been reduced from 10% to 0.4% (23). Spasticity and hip displacement without dislocation could be an explanation for the hip pain reported. In general, pain in the lower limbs seems to be the most common location for pain in CP (3,4). We also found that those in GMFCS V had more pain in the abdomen than those at GMFCS I, II and III. This might be caused by constipation or gastroesophageal reflux, which have been found to be more frequent among those in GMFCS V (24). In other large population-based studies on older children, headaches seem to have been more frequent than pain in the abdomen (6), which was also in accordance with reports on children and adolescents who did not have CP (19). Except for the lower extremities, there seem to be no clear patterns in terms of where individuals with CP hurt. Parkinson et al. found that those in GMFCS level III were more likely to have pain in multiple sites (6). We also noticed this pattern in our study, but the findings were not statistically significant.

There were a number of limitations to our study. Only one generic screening item was available to assess the presence of pain, which meant that we were not able to assess the severity, duration or type of pain. This information would have allowed us to provide a more complete picture of pain in children and adolescents with CP. Although self-reports are considered the gold standard, there are times when proxy reports are necessary. The use of proxies, and our inability to determine when proxies were in fact used, can be viewed as limitations. Communication and intellectual difficulties are more common in GMFCS V, and therefore, proxy reports occurred disproportionally more often for those individuals. In our case, proxy reports were also more likely when the children were infants and toddlers. It is not well established how proxy and self-reports of pain are correlated and both over and under reports have been hypothesised (3,25). Changes will be made to CPUP in the future to include data on the use of proxies.

There are many questions that remain in terms of pain in CP. To the best of our knowledge, there have been no descriptive or explanatory studies on trajectories of pain over time in a total population. Finally, even though prevalence of pain may differ among studies for different reasons, it seems clear that a sufficient body of evidence has emerged to conclude that pain is a significant challenge for individuals with CP and that current treatment or pain management plans might be insufficient. Concerted research needs to be conducted to determine what can be done to reduce the high frequency of pain in this population.

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CONFLICTS OF INTEREST

The authors have no conflict of interests to declare.

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