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REGULAR ARTICLE

Pain in children with cerebral palsy: a cross-sectional multicentre European study

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Abstract

Aim: To determine the prevalence and associations of self-reported and parent-reported pain in children with cerebral palsy (CP) of all severities.

Method: Cross-sectional design using a questionnaire; analysis using ordinal regression. Children aged 8–12 years were randomly selected from population-based registers of children with CP in eight European regions; a further region recruited 75 children from multiple sources. Outcome measures were pain in the previous week among children who could self-report and parents' perception of their child's pain in the previous 4 weeks.

Results: Data on pain were available from 490 children who could self-report and parents of 806 children (those who could and could not self-report). The estimated population prevalence of self-reported pain in the previous week was 60% (95% CI: 54–65%) and that of parent-reported pain in the previous 4 weeks was 73% (95% CI: 69–76%). In self-reporting children, older children reported more pain but pain was not significantly associated with severity of impairment. In parent reports, severity of child impairment, seizures and parental unemployment were associated with more frequent and severe pain.

Conclusion: Pain in children with CP is common. Clinicians should enquire about pain and consider appropriate physical, therapeutic or psychological management.

INTRODUCTION

Pain is common in children with neuromuscular and neurodevelopmental problems such as cerebral palsy (CP). Chronic pain is frequent in children with moderate to severe CP (1). It is related to mobility and other demanding activities, and interferes with sleep (2). Children with CP who have pain participate less in everyday situations (1,3) and experience lower quality of life than those without pain (4).

Using age appropriate instruments, children should report their own pain whenever possible. This may be difficult for children with CP because some have speech impairment, which makes it difficult to communicate their experience (5,6) and others have such severe cognitive impairment that proxy reports are necessary (7).

The aim of this study was to report the experience of pain in a large sample of children with CP within a narrow age range and across the whole spectrum of severity. We describe pain in the children who can self-report. In addition, we describe parent reports for all children. We examine the prevalence of the pain and its associations with the child's impairments and with a range of socio-demographic characteristics.

METHODS

Participants

This study is part of a wider project (SPARCLE) and its methods have been reported in detail (8). Eligible children were born between 31st July 1991 and 1st April 1997 and held on registers of children with CP covering eight regions of six European countries (9). A total of 1174 families were sampled and 743 (63%) agreed to participate (10). A further region recruited 75 children from multiple sources. Thus, 818 children comprised the sample. Trained research associates visited the families at home, if possible when the children were aged between 8 and 12 years old. Parents were asked if their child would be able to understand a questionnaire. If this was unclear, their child's understanding of a scale was assessed by a test requiring the child to order cubes of different sizes (11).

Measures

The two items from the 'Bodily Pain and Discomfort' scale of the Child Health Questionnaire (12) were used to capture pain. Each item asks about pain experienced over the previous 4 weeks, one focusing on frequency and the other on severity. Parents reported their child's pain so that we had a common metric for all children. Where possible, we sought a child's own report of pain over the past week.

Parents provided information about their own background, family circumstances, and their child's age, gender, number and disability of siblings, school type and impairments, gross motor function using the Gross Motor Function Classification System (GMFCS) (13), hand function using the Bimanual Fine Motor Function system (14), seizures, feeding, communication, vision and hearing. Child

IQ was estimated by asking parents about the child's understanding, learning and friendships. CP type was available from the registers.

Signed consent was sought from all parents. Children deemed capable of self-completion were invited to give their own consent to take part in the study. Ethics Committee approval was obtained in each country.

Statistical methods

We treated the six possible responses for frequency and severity of pain as ordinal variables and used proportional odds ordinal regression (15) to relate them to the child's impairments and socio-demographic characteristics. These models allowed us to estimate odds ratios (ORs) and their 95% confidence intervals comparing the odds of pain among children in a specific group (e.g. defined by type and severity of impairment) with the odds of pain in a reference group (e.g. the least impaired children). The model assumes that the ORs comparing pain in two groups of children (e.g. severely and mildly impaired) are the same irrespective of how the six categories of pain are dichotomized. For example, we can define the odds of pain as response in categories 2–6 vs. category 1, or categories 3–6 vs. categories 1–2, etc.

All models were stratified by region. Univariate analyses were performed, relating frequency and severity of pain to each type of impairment and each socio-demographic characteristic in turn. Forwards stepwise regression, followed by a backwards step, was then used to select covariates to include in a multivariable model. The criteria for inclusion were based on the p-value from the likelihood ratio test statistic (15). To lessen the probability of chance findings because of multiple hypotheses testing, the p-value for entry and removal of covariates was set at 0.01. Finally, the penultimate models were re-estimated using weights that allowed for the sampling design and non-response (10). This allowed us to estimate, from our sample, the prevalence of pain in the population of all children with CP.

Goodness-of-fit of the final multivariable models was assessed by calculating a fit statistic comparing the observed and expected number of children with pain in 10 quantiles of risk of pain (16). Sensitivity analyses were performed excluding children with the largest residuals. Statistical analyses were performed using the Stata 10 (Stata Corporation, College Station, TX, USA).

RESULTS

Parent-reported pain was available for 806 children and 490 children reported their own pain. Eight children had missing data on some aspects of impairment. The different types of impairment were strongly correlated: the Spearman rank correlation between all pairs of types of impairment (excluding vision and hearing) were large (0.33–0.75) and highly significant ($p < 0.0001$). Self-reporting children tended to be less severely impaired than other children in terms of walking ability, bimanual fine motor function, feeding and communication ability and IQ (Table S1).

Figure 1 summarizes the frequency and severity of self-reported and parent-reported pain, both overall and by walking ability. Although self-reported pain varied little with walking ability, parents tended to report more frequent and more severe pain if their child was more severely impaired, in particular if the child could not walk. In our sample, 56% of self-reporting children experienced some pain in the previous week and 72% of parents (i.e. of children who could and could not self-report) said that their child had some pain in the previous 4 weeks. After allowing for the sampling design and for non-response, these proportions corresponded to population prevalences of self-reported pain of 60% (95% CI: 54–65%) among children who could self-report and of parent-reported pain of 73% (95% CI: 69–76%) among all children.

Regression model: self-reported pain in the previous week

In the univariable analysis, the frequency and severity of self-reported pain did not vary significantly with impairment, but younger children and those living in a village or the countryside tended to report less frequent and less severe pain. As younger children were more likely than older children to live in a village or the countryside, only frequency of pain remained significantly ($p < 0.01$) associated with age in multivariable analysis (Table 1).

Regression model: parent-reported pain in the previous 4 weeks

In the univariable analysis, parents tended to report a higher frequency and severity of pain if the child was more severely impaired in terms of walking ability, hand function, seizures, feeding and communication difficulties, intellect and CP type or if the child was a girl or attended a special school or a special unit in a mainstream school or if only one parent worked part-time or neither parent worked (Table S1). In multivariable analysis (Table 1), only walking ability, seizures and parental unemployment were significantly associated with frequency of pain. For severity of pain, our scheme for choosing variables for the multivariable model selected fine motor skills, seizures and parental unemployment; however, a model with walking ability instead of fine motor skills was just as satisfactory in terms of the likelihood, and we preferred the latter model as it was similar to the model for frequency of pain.

For all final multivariable models, sensitivity analyses excluding children with the largest residuals yielded similar results; and goodness-of-fit statistics were satisfactory ($p > 0.10$; Table 1).

DISCUSSION

Main findings and comparison with other studies

Pain is a common experience for children with CP. Extrapolating from our sample to the wider population of children with CP, we estimated that 60% (95% CI: 54–65%) of children able to report their own pain have experienced some pain in the previous week. This is comparable with the findings of Engel et al. (2) who estimated 70% prevalence in

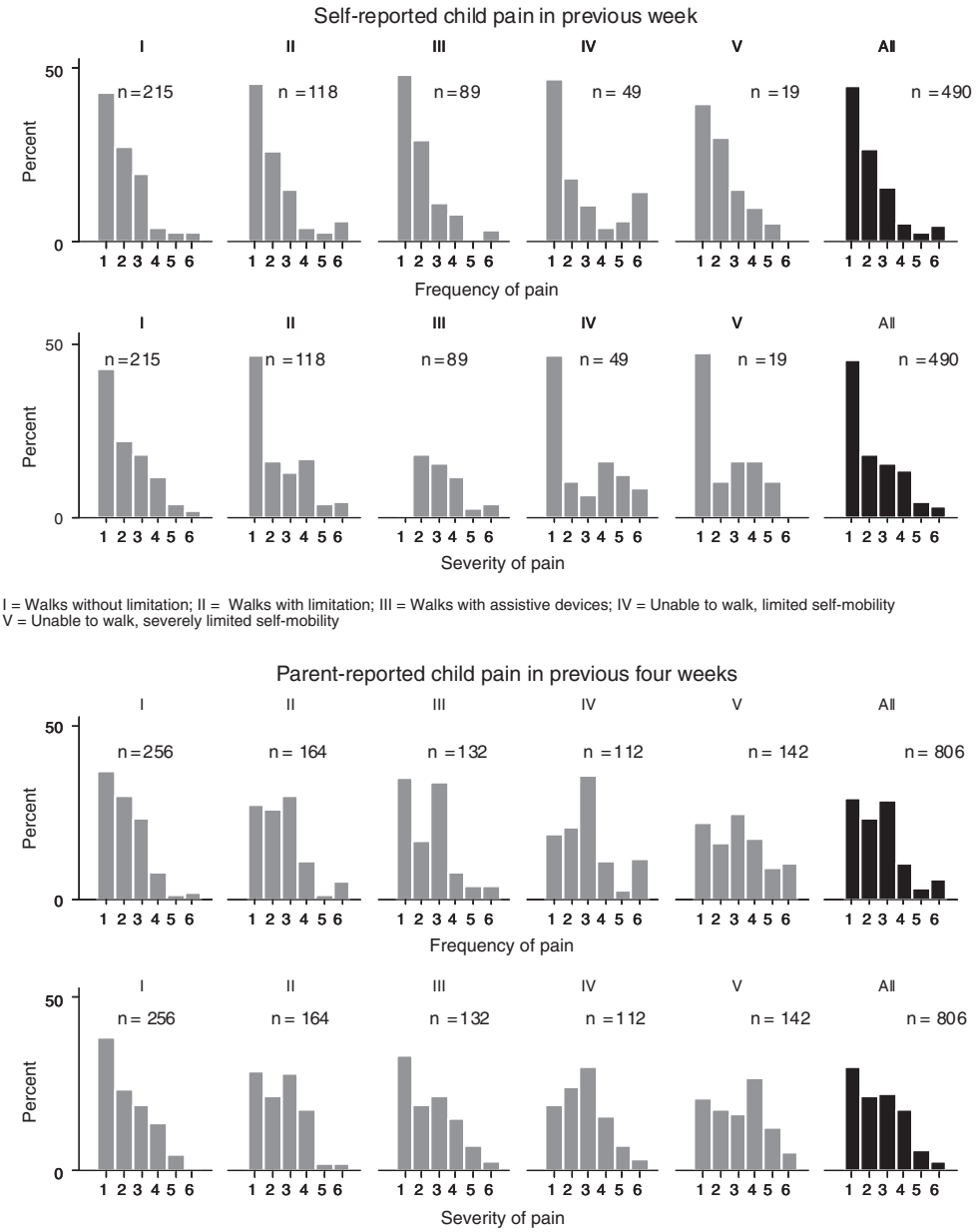

Figure 1 Distribution of self-reported pain over 1 week and parent-reported pain over 4 weeks, overall and by level of walking ability. For frequency of pain: 1 = never; 2 = once or twice; 3 = a few times; 4 = fairly often; 5 = very often; 6 = every day or almost every day. For severity of pain: 1 = none; 2 = very mild; 3 = mild; 4 = moderate; 5 = severe; 6 = very severe.

6- to 17-year-old children with CP. Both are much higher than that of self-reporting children of a similar age in the general population, estimated as 35% (17) and 33% (18). Seventy-three per cent (95% CI: 69–76%) of parents of all children (those who can and cannot self-report) perceive their child to have experienced some pain in the previous 4 weeks. We found only one other study of parent-reported pain in the previous month in children with CP of all severities and it reported a prevalence of 14% (19). However, it excluded children who had received certain treatments and used the Health Utilities Index, which only asks about pain that restricts activities.

Among self-reporting children, pain was not associated with impairment. Older children tended to report more pain than younger children, consistent with previous studies (17,18). In parent reports of pain for the whole sample, more frequent and more severe pain was associated with more severely impaired motor function, seizures and parental unemployment, but was not associated with age. Despite undertaking a number of *post hoc* analyses, it was unclear

Table 1 Odds ratios (OR) from multivariable ordinal regression models, relating child pain to the type and level of impairment and socio-demographic characteristics

	Children who could self-report (n = 490) Self-reported pain in previous week					All children (n = 799): parent-reported pain in previous 4 weeks					
	OR*	Frequency (95% CI)	p†	OR*	Severity (95% CI)	OR*	Frequency (95% CI)	p†	OR*	Severity (95% CI)	p†
GMFCS											
I. Walks without limitation						1.0		<0.0001	1.0		<0.0001
II. Walks with limitation						1.5	1.0–2.3		1.3	0.9–2.0	
III. Walks with assistive devices						1.2	0.7–1.9		1.2	0.7–1.9	
IV. Unable to walk, limited self-mobility						2.0	1.2–3.3		1.6	1.0–2.6	
V. Unable to walk, severely limited self-mobility						2.8	1.6–4.7		3.0	1.8–5.0	
Seizures (in previous year)											
No, Seizures, not on medication						1.0		0.0008	1.0		0.0003
No, Seizures, on medication						0.8	0.5–1.5		0.7	0.4–1.1	
Seizures less than once a month						1.6	0.8–3.1		1.6	0.9–2.9	
Seizures between once a month & once a week						2.0	1.1–3.8		2.4	1.2–4.5	
Seizures more than once a week						2.4	1.3–4.2		2.1	1.1–4.0	
Age in years											
7–8	1.0		0.007								
9	1.0	0.5–2.0									
10	1.8	1.0–3.2									
11	2.2	1.2–3.9									
12–13	2.2	1.2–4.0									
Parental employment											
At least one parent works full-time professionally (or equivalent)						1.0		0.002	1.0		0.003
Intermediate						0.8	0.6–1.2		1.0	0.6–1.4	
One parent works part-time or neither parent works						1.8	1.1–2.9		1.6	1.1–2.5	
p-value from goodness-of-fit deciles of risk test	0.32					0.76			0.55		

All models were stratified by region and used weights that allowed for the sampling design and for non response. Children with missing data, either on pain outcomes or on factors included in the model, were excluded.

*ORs greater than 1.0 indicate a higher level of pain in that group than in the reference group.

†p-values are from likelihood ratio test statistic comparing models without the corresponding factor, and these models did not use sampling weights.

whether the lack of significant association between self-reported pain and impairment – in contrast to the significant association between parent-reported pain and impairment – was due to the smaller number of self-reporting children and the limited variability in their level of impairment or to real differences between children and parents in their perception of child pain.

The association between the severity of motor impairment and parent-reported pain is, however, consistent with previous studies (1,20). This association is likely to be causal because persons with more severe impairment tend to have more spasticity, which causes pain and needs painful treatments. The association between parent-reported pain and seizures is consistent with a study by Breau et al. (21). Another study found no such association (20), although this might be because it included children with both well- and poorly controlled epilepsy. Epilepsy does not cause pain but is a marker of wider brain damage, often associated with swallowing difficulties and gastro-oesophageal reflux. The association between pain and parental unemployment could be partly because parents are less likely to work when caring for a child with complex needs and chronic pain or because parental unemployment may increase parental stress, which in turn may heighten parents' perception of their child's pain. A similar association between pain and low income (22) has been reported in the general population.

Strengths and limitations of the study

Participants were selected from children with all levels of severity of CP in nine geographic areas, eight of which had population-based registers, ensuring a representative sample. Although non-response was 37%, the weights allowed for the pattern of non-response, which should reduce bias (10) but cannot eliminate bias if the experience of pain was systematically different from that of responders.

The study had a large sample of children in a narrow age range. A standardized protocol was used to assess cognitive ability. The pain questions in the CHQ were well validated. We retained the ordinal categories as recommended (23) and then used ordinal regression, thus having more information and therefore more statistical power.

Implications for clinical practice

It is clear that pain is common in children with CP. By increasing clinicians' awareness on the high prevalence of pain in this group, we can promote earlier identification, better investigation of the causes and better management of pain in children with CP. Self-report of pain by the child is the ideal. Parent proxy report is appropriate for some children, and there is a validated assessment tool for children with severe communication difficulty, which is potentially useful clinically (24). Better management can start by asking children and their parents about pain as children with CP may always have had pain and assume it to be normal. Discussion of such pain is itself helpful (25). Analgesics and local anaesthetic should be used when painful procedures are anticipated. Psychological factors play an important part in most chronic pain. Cognitive behaviour therapy – used for adults with CP (26) – may also be helpful for children.

Implications for research

Pain is common in adults with CP and in particular greater pain is associated with deterioration in walking function, fatigue and balance (27). It is possible that a predisposition to adult pain is established in childhood either through damage to joints and back or through conditioning because of chronic unrecognized and untreated pain. Recent developments in the understanding of pain suggest that the brain may have a 'memory' for pain, which maintains pain at levels unrelated to the current noxious stimulus (28). About a half of children with CP need neonatal intensive care and the effects of painful stimuli on immature neural pain circuits are now known to influence subsequent pain processing (28). While animal models suggest that increased or reduced sensitivity to pain may be induced, a clinical study suggests that adolescents born preterm have increased sensitivity (29). We did not find any study on the neurobiological development of pain pathways in the damaged brain and this could be an important area for research.

Better understanding is also needed of the types and immediate causes of pain due to the CP itself. Pain related to CP includes: chronic pain such as from operation sites, spasms, deformity, dislocation, capsulitis and hips; acute pain such as gastro-oesophageal reflux; and procedural pain from botulinum therapy, operations and therapy. In one study, assisted stretching was the daily activity most frequently identified as painful (30), which highlights the need to record adverse effects of treatment and balance them against the benefits.

Epidemiological studies are needed to establish if pain is reported to clinicians and, if so, whether it is actively managed. In addition, prospective evaluations using pain diaries and randomized controlled trials of interventions are needed.

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SUPPORTING INFORMATION

Additional Supporting Information may be found in the online version of this article:

Table S1 Odds ratios (OR) from univariable ordinal regression models, relating child pain to impairment and socio-demographic characteristics

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