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Pain in young people aged 13 to 17 years with cerebral palsy: cross-sectional, multicentre European study

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ABSTRACT

Objective To determine the prevalence and associations of self- and parent-reported pain in young people with cerebral palsy (CP).

Design and setting Cross-sectional questionnaire survey conducted at home visits in nine regions in seven European countries. Participants were 13 to 17-year-olds (n=667) drawn from population CP registers in eight regions and from multiple sources in one region. 429 could self-report; parent-reports were obtained for 657. Data were collected on: severity, frequency, site and circumstances of pain in previous week; severity of pain associated with therapy in previous year.

Results The estimated population prevalence of any pain in previous week was 74% (95% CI 69% to 79%) for self-reported pain and 77% (95% CI 73% to 81%) for parent-reported pain. 40% experienced leg pains, 34% reported headaches and 45% of those who received physiotherapy experienced pain during therapy. Girls reported more pain than boys (OR=2.1, 95% CI 1.5 to 3.0) and young people reported more pain if they had emotional difficulties (comparing highest and lowest quartiles: OR=3.1, 95% CI 1.7 to 5.6). Parents reported more pain in children with emotional difficulties (OR=4.2, 95% CI 2.7 to 6.6), or with more impaired walking ability.

Conclusions Pain in young people with CP is highly prevalent. Because pain causes immediate distress and is associated with lower subjective well-being and reduced participation, clinicians should routinely assess pain. Clinical interventions to reduce pain should be implemented and evaluated. The efficacy of medical and therapeutic interventions causing pain should be re-examined to establish if their benefit justifies the pain and fear of pain that accompany them.

INTRODUCTION

The literature on pain in adolescents with cerebral palsy (CP) is small but there have been two recent studies. The first¹ reported pain in a population-based sample across the severity spectrum from young people's self-reports where possible and otherwise from parents' reports; analyses combined these as if they were equivalent. The second² reported pain from one sample derived from organisations that work with young people with CP and one from a hospital-based group; results may therefore be unrepresentative of young people with CP.

In a large European Study, SPARCLE (Study of PARTICipation of children with Cerebral palsy Living in Europe),³ of 8 to 12-year-old children with CP, pain was very prevalent⁴ and was consistently associated with lower subjective well-being⁵ and reduced participation.⁶ Therefore when the

What is already known on this topic

- ▶ Pain in young people with cerebral palsy is associated with lower subjective well-being and reduced participation.
- ▶ There are many potential causes of pain in cerebral palsy such as spasms, hip subluxation, contractures, operation sites, therapeutic interventions, assistive devices and gastrostomy tubes.

What this study adds

- ▶ Pain in young people with cerebral palsy is very prevalent: about 75% experience some pain in a typical week. Strategies to reduce pain appear to be absent or inadequate.
- ▶ Clinicians should routinely assess pain and develop pain management plans if necessary.
- ▶ Much pain is caused by clinical procedures or therapies; their efficacy should be re-examined to establish if they deliver sufficient benefit to justify the pain and fear of pain that accompany them.

young people were visited again aged 13 to 17 years (SPARCLE2),⁷ we collected more detailed information about pain.

In this paper we report self- and parent-reported pain, and analyse these separately. We report prevalence of pain, sites and circumstances of pain, and examine the associations of pain with young people's impairments and emotional difficulties, parenting stress and sociodemographic characteristics. We also compare self-reports of pain with parents' reports of their child's pain.

METHODS

The methods of the SPARCLE studies have been described in detail elsewhere^{3 7-9} and are summarised briefly below.

Participants

SPARCLE1 randomly sampled 1174 children from population-based registers of children with CP. Children were eligible if born between 31 July 1991 and 1 April 1997. The registers cover eight regions of six European countries (table 1) that share a standardised definition and classification of CP.¹⁰ One

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Table 1 Distribution of impairments, sociodemographic characteristics, pain, emotional difficulties score and total stress score

| | Self-reported (n=429) | | Parent-reported (n=657) | |
|---|--------------------------|-------|----------------------------|-------|
| | n | (%) | n | (%) |
| <i>Impairments of young people</i> | | | | |
| Walking ability as captured by gross motor function | | | | |
| I. Walks without limitation | 199 | (46%) | 226 | (34%) |
| II. Walks with limitation | 84 | (20%) | 113 | (17%) |
| III. Walks with assistive devices | 58 | (14%) | 85 | (13%) |
| IV. Unable to walk, limited self-mobility | 50 | (12%) | 90 | (14%) |
| V. Unable to walk, severely limited self-mobility | 37 | (9%) | 143 | (22%) |
| Information not available | 1 | (0%) | 0 | (0%) |
| Seizures | | | | |
| No seizures, not on medication | 367 | (86%) | 464 | (71%) |
| No seizures, on medication | 23 | (5%) | 73 | (11%) |
| Seizures less than once a month | 19 | (4%) | 46 | (7%) |
| Seizures between once a month and once a week | 9 | (2%) | 24 | (4%) |
| Seizures more than once a week | 6 | (1%) | 45 | (7%) |
| Information not available | 5 | (1%) | 5 | (1%) |
| Intellectual ability | | | | |
| IQ > 70 | 286 | (67%) | 300 | (46%) |
| IQ 50–70 | 129 | (30%) | 169 | (26%) |
| IQ < 50 | 13 | (3%) | 187 | (28%) |
| Information not available | 1 | (0%) | 1 | (0%) |
| Cerebral palsy subtype | | | | |
| Unilateral spastic | 175 | (41%) | 208 | (32%) |
| Bilateral spastic | 208 | (48%) | 354 | (54%) |
| Dyskinetic | 28 | (7%) | 62 | (9%) |
| Ataxic | 13 | (3%) | 28 | (4%) |
| Information not available | 5 | (1%) | 5 | (1%) |
| <i>Sociodemographic characteristics</i> | | | | |
| Regions | | | | |
| Southeast France | 41 | (10%) | 61 | (9%) |
| Southwest France | 38 | (9%) | 57 | (9%) |
| Southwest Ireland | 52 | (12%) | 76 | (12%) |
| West Sweden | 40 | (9%) | 66 | (10%) |
| North England | 73 | (17%) | 107 | (16%) |
| Northern Ireland | 64 | (15%) | 88 | (13%) |
| East Denmark | 53 | (12%) | 86 | (13%) |
| Central Italy | 17 | (4%) | 42 | (6%) |
| Northwest Germany | 51 | (12%) | 74 | (11%) |
| Gender | | | | |
| Boys | 249 | (58%) | 376 | (57%) |
| Girls | 180 | (42%) | 281 | (43%) |
| Age in years | | | | |
| <13 | 27 | (6%) | 45 | (7%) |
| 13 | 99 | (23%) | 137 | (21%) |
| 14 | 85 | (20%) | 135 | (21%) |
| 15 | 91 | (21%) | 131 | (20%) |
| 16 | 76 | (18%) | 114 | (17%) |
| 17 | 47 | (11%) | 83 | (13%) |
| >17 | 4 | (1%) | 12 | (2%) |
| Information not available | 5 | (1%) | 9 | (1%) |
| <i>Pain</i> | | | | |
| Frequency of pain in previous week | | | | |
| None of the time | 133 | (31%) | 180 | (27%) |
| Once or twice | 143 | (33%) | 161 | (25%) |
| A few times | 79 | (18%) | 140 | (21%) |
| Fairly often | 29 | (7%) | 64 | (10%) |
| Very often | 16 | (4%) | 38 | (6%) |
| Every day | 29 | (7%) | 74 | (11%) |

Continued

Table 1 Continued

| | Self-reported (n=429) | | Parent-reported (n=657) | |
|---|--------------------------|-------|----------------------------|-------|
| | n | (%) | n | (%) |
| Severity of pain in previous week | | | | |
| None | 130 | (30%) | 180 | (27%) |
| Very mild | 94 | (22%) | 100 | (15%) |
| Mild | 96 | (22%) | 133 | (20%) |
| Moderate | 64 | (15%) | 176 | (27%) |
| Severe | 26 | (6%) | 55 | (8%) |
| Very severe | 19 | (4%) | 13 | (2%) |
| Emotional difficulties score (by quartile)* | | | | |
| 0–1 | 98 | (23%) | 179 | (27%) |
| >1–3 | 146 | (34%) | 206 | (31%) |
| >3–5 | 109 | (25%) | 164 | (25%) |
| >5–10 | 72 | (17%) | 106 | (16%) |
| Information not available | 4 | (1%) | 2 | (0%) |
| Total stress score (by quartile)† | | | | |
| 36–64 | 127 | (30%) | 163 | (25%) |
| 65–80 | 118 | (28%) | 162 | (25%) |
| 81–97 | 92 | (21%) | 167 | (25%) |
| 98–147 | 81 | (19%) | 152 | (23%) |
| Information not available | 11 | (3%) | 13 | (2%) |

*Higher scores indicate more emotional difficulties.

†Higher scores indicate more stress.

further region (northwest Germany) ascertained 75 cases from multiple sources. The 818 children who entered SPARCLE1 were followed up when aged 13 to 17 years; 594 (73%) agreed to participate. In order to maintain statistical power for cross-sectional analyses,^{7–9} SPARCLE2 additionally sampled from young people eligible for SPARCLE1 who had not participated in it. Seventy-three agreed to participate and hence the final sample for SPARCLE2 comprised 667 young people, distributed by region as shown in table 1.

Procedure

Researchers visited families in their homes, if possible when the young people were aged 13 to 17 years. Parents were asked to report on the measures below for all the young people. Young people who could self-report were asked to report their pain.

Measures

- Any pain, measured using the Bodily Pain and Discomfort items of the Child Health Questionnaire.¹¹ The items are valid and reliable^{12–13} and record frequency of pain (none of the time, once or twice, a few times, fairly often, very often, every day) and severity (none, very mild, mild, moderate, severe, very severe); we changed the timeframe to 1 week to accord with the wider SPARCLE study.
- Site and circumstances of pain, as shown in the left hand column of table 2, recording frequency and severity in the previous week using the response categories above.
- Severity of pain during treatment over the previous year (table 2) using the response categories above.
- Emotional difficulties score (EDS) from the Strengths and Difficulties Questionnaire.^{14–15}
- Parenting stress using the total stress score from the Parenting Stress Index Short Form.¹⁶

- Impairment: walking ability described by gross motor function¹⁷; fine motor function¹⁸; seizures; feeding; communication; intellectual ability¹⁹; and CP type.
- Sociodemographic characteristics: parents' employment and educational qualifications, family structure, area of domicile, child's school type.

Statistical methods

To estimate the population prevalence of any pain, we dichotomised severity of pain as none/any (from very mild to very severe) and restricted the sample to the young people aged 13 to 17 in SPARCLE2 who had participated in SPARCLE1 and for whom sampling weights were therefore available; young people in northwest Germany were excluded as this region did not sample from population-based registers.

For all other statistical analysis, pain was not dichotomised; we used proportional odds ordinal regression which retained all six categories of severity and frequency of pain.²⁰ We modelled the association between pain and covariates (impairments, socio-demographic characteristics, EDS, total stress score), stratifying by region. For analysis of trend, walking ability was treated as continuous; for all other analyses, covariates were treated as categorical. Four models, corresponding to young people's and parents' responses were developed. We first performed univariable analyses, relating pain to each covariate in turn. We then used forwards stepwise regression, followed by backwards steps, to select covariates to include in a multivariable model. We set the p value for entry of covariates as $p < 0.05$ and, to lessen the probability of chance findings due to multiple hypotheses testing, we set the p value for removal of covariates at 0.01. We derived p values from the likelihood ratio test statistic. We checked for an interaction between significant covariates. We performed sensitivity analyses: (a) limiting the sample to young people who had responded to SPARCLE1 and for whom

Table 2 Site and circumstances of pain severity in previous week, by walking ability*

| | (a) Self-report of pain by young people | | | | | | | | (b) Parent-report of their child's pain | | | | | | | |
|--|---|----|----|----|-----|----|----|---------|---|----|----|----|-----|----|----|---------|
| | Walking ability (GMFCS) | | | | | | | | Walking ability (GMFCS) | | | | | | | |
| | All | | I | II | III | IV | V | p Value | All | | I | II | III | IV | V | p Value |
| | N | % | % | % | % | % | % | | N | % | % | % | % | % | % | |
| Site of pain (in previous week) | | | | | | | | | | | | | | | | |
| Headaches | 423 | 34 | 39 | 38 | 36 | 18 | 19 | 0.004 | 629 | 30 | 33 | 40 | 24 | 31 | 20 | 0.002 |
| Stomach | 422 | 26 | 26 | 30 | 25 | 24 | 27 | 0.49 | 634 | 32 | 24 | 33 | 23 | 42 | 44 | <0.001 |
| Back | 420 | 27 | 27 | 24 | 28 | 22 | 41 | 0.12 | 632 | 25 | 19 | 20 | 23 | 31 | 38 | <0.001 |
| Arms | 416 | 18 | 19 | 20 | 14 | 12 | 17 | 0.10 | 630 | 14 | 9 | 17 | 13 | 13 | 22 | 0.020 |
| Hips | 421 | 14 | 13 | 12 | 14 | 18 | 22 | 0.07 | 635 | 21 | 9 | 16 | 17 | 28 | 44 | <0.001 |
| Legs | 413 | 40 | 39 | 41 | 47 | 40 | 37 | 0.19 | 634 | 43 | 37 | 48 | 49 | 48 | 41 | 0.07 |
| Operation sites | 411 | 10 | 5 | 12 | 14 | 16 | 17 | <0.001 | 624 | 14 | 5 | 16 | 21 | 19 | 21 | <0.001 |
| Circumstances of pain (in previous week) | | | | | | | | | | | | | | | | |
| At rest from spasms | 413 | 13 | 10 | 13 | 16 | 20 | 14 | 0.04 | 632 | 17 | 8 | 16 | 16 | 18 | 35 | <0.001 |
| At rest, from splints or restraints | 410 | 12 | 8 | 13 | 21 | 18 | 6 | 0.03 | 625 | 20 | 9 | 19 | 25 | 31 | 29 | <0.001 |
| On swallowing or feeding | 410 | 3 | 2 | 3 | 4 | 4 | 6 | 0.12 | 627 | 6 | 4 | 3 | 1 | 11 | 11 | <0.001 |
| On moving | 414 | 28 | 19 | 36 | 35 | 29 | 42 | <0.001 | 634 | 35 | 19 | 35 | 45 | 52 | 42 | <0.001 |
| On changing or dressing | 413 | 5 | 2 | 3 | 4 | 8 | 19 | <0.001 | 627 | 15 | 4 | 6 | 12 | 26 | 38 | <0.001 |
| In bed at night | 415 | 17 | 16 | 14 | 11 | 22 | 32 | 0.02 | 633 | 22 | 11 | 15 | 23 | 32 | 38 | <0.001 |
| Short unexpected pains | 413 | 30 | 27 | 41 | 28 | 22 | 33 | 0.25 | 627 | 25 | 19 | 24 | 22 | 29 | 36 | <0.001 |
| Pain during therapy (in previous year) | | | | | | | | | | | | | | | | |
| During physiotherapy | 339 | 45 | 38 | 42 | 50 | 62 | 50 | 0.002 | 505 | 50 | 40 | 51 | 46 | 58 | 60 | <0.001 |
| During other therapy | 160 | 9 | 4 | 11 | 14 | 17 | 6 | 0.32 | 281 | 18 | 8 | 17 | 17 | 22 | 29 | 0.003 |
| During botulinum injections | 142 | 26 | 22 | 28 | 35 | 21 | 36 | 0.42 | 233 | 29 | 16 | 33 | 44 | 34 | 29 | 0.32 |

p, Significance of trend of pain over levels of walking ability, estimated using ordinal regression which retained all six categories of pain.

N, Number included: data were unavailable if Gross Motor Function Classification System (GMFCS) or severity of pain were not reported or if the young person had not received the relevant therapy.

%, Percentage with severity of pain from very mild to very severe.

*Presence of pain was defined by severity in any category from very mild to very severe.

sampling weights that reflected the sampling design were available; and (b) retaining the entire sample but additionally adjusting for factors associated with non-response.^{8 9 21}

Stata V.12 was used for analysis.

Ethics

Ethics approval was obtained or a statement that only registration was required as appropriate in each country. Signed consent was obtained from all parents and from young people who could give meaningful consent.

RESULTS

Of the 667 young people in SPARCLE2, 429 (64%) reported their own pain; parents' reports of their child's pain were available for 657 (99%). The distributions of some impairments, some sociodemographic variables, pain, EDS and parenting stress score are presented in table 1. The Spearman rank correlations between all pairs of types of impairment (except CP type) were strong ($\rho=0.24$ to 0.75) and statistically significant ($p<0.0001$). About one third of the young people reported no pain in the previous week, a third reported pain once or twice, and a third reported more frequent pain.

Prevalence of pain

Based on severity, the proportion of young people with any pain in the previous week was 70% by self-report and 73% by parent-report. Using the prevalence sample, these proportions corresponded to population prevalences of self-reported pain of 74% (95% CI 69% to 79%) and of parent-reported pain of

77% (95% CI 73% to 81%). Results based on frequency of pain were similar.

Sites and circumstances of pain

The percentages of young people reporting any pain in the previous week at specific sites and in specific circumstances are presented in table 2. The most common sites for pain were legs (40%) and head (34%). Over a quarter reported stomach or back pain, pain on moving or short unexpected pains. Those with more severe impairment of walking ability were significantly more likely to report pain on moving, changing or dressing, or at operation sites. Those with less severe impairment of walking ability tended to report more headaches.

The distribution of parent-reported pain was similar, although trends over walking ability were more marked. Parents perceived their child as experiencing more pain in the stomach, back and hips, and at operation sites, and more pain in all the circumstances considered if they had more impaired walking ability. Results based on frequency of pain were similar.

Pain during therapy

Almost half of young people who could self-report and who had received physiotherapy in the previous year reported pain during therapy; 30% reported very mild or mild pain, 9% moderate pain and 6% severe or very severe pain. Pain during physiotherapy was significantly more likely to occur if the young person had more severely impaired walking ability (table 2). Over a quarter of young people reported pain during botulinum injections; 11% reported very mild or mild pain, 4%

Table 3 ORs from multivariable ordinal regression models of young people's pain in previous week*

| | Severity of pain | | | Frequency of pain | | |
|--|------------------|--------------|----------|-------------------|--------------|----------|
| | OR† | (95% CI) | p Value‡ | OR† | (95% CI) | p Value‡ |
| <i>Self-report of pain by young person (n=425)</i> | | | | | | |
| Gender | | | <0.0001 | | | 0.0006 |
| Boy | 1.0 | | | 1.0 | | |
| Girl | 2.1 | (1.5 to 3.0) | | 1.9 | (1.3 to 2.7) | |
| Emotional difficulties score (by quartile) | | | 0.001 | | | 0.0002 |
| 0–1 | 1.0 | | | 1.0 | | |
| >1–3 | 1.4 | (0.9 to 2.3) | | 1.3 | (0.8 to 2.2) | |
| >3–5 | 1.8 | (1.1 to 3.0) | | 1.8 | (1.1 to 3.1) | |
| >5–10 | 3.1 | (1.7 to 5.6) | | 3.5 | (1.9 to 6.3) | |
| <i>Parent-report of their child's pain (n=655)</i> | | | | | | |
| Emotional difficulties score (by quartile) | | | <0.0001 | | | <0.0001 |
| 0–1 | 1.0 | | | 1.0 | | |
| >1–3 | 1.7 | (1.2 to 2.4) | | 1.5 | (1.1 to 2.2) | |
| >3–5 | 2.4 | (1.6 to 3.5) | | 2.0 | (1.4 to 3.0) | |
| >5–10 | 4.2 | (2.7 to 6.6) | | 4.1 | (2.6 to 6.4) | |
| Walking ability (captured by gross motor function) | | | <0.0001 | | | <0.0001 |
| I. Walks without limitation | 1.0 | | | 1.0 | | |
| II. Walks with limitation | 1.4 | (1.0 to 2.2) | | 1.3 | (0.9 to 2.0) | |
| III. Walks with assistive devices | 1.5 | (0.9 to 2.4) | | 1.6 | (1.0 to 2.5) | |
| IV. Unable to walk, limited self-mobility | 2.1 | (1.3 to 3.2) | | 2.0 | (1.3 to 3.0) | |
| V. Unable to walk, severely limited self-mobility | 5.1 | (3.4 to 7.6) | | 4.9 | (3.3 to 7.4) | |

*All models were stratified by region. Young people with missing data, either on pain outcomes or on factors included in the model, were excluded.

†ORs <1.0 indicate a higher level of pain in that group than in the reference group.

‡p Values are from the likelihood ratio test statistic comparing models with and without the corresponding factor.

moderate pain and 11% severe or very severe pain. Parents' reports confirmed these findings.

Associations of pain

In univariable analysis of any self-reported pain (as captured by the instrument described in Measures (1) in the Methods section), the severity did not vary significantly ($p < 0.01$) with any type of impairment or with any sociodemographic characteristic except young people's gender: girls tended to report more severe pain. Young people with a higher EDS also tended to report more severe pain. Both gender and EDS remained statistically significant in a multivariable model (table 3). Results for frequency of pain were similar. Sensitivity analyses yielded similar results.

In univariable analysis of parents' reports of their child's pain, pain was significantly more frequent and more severe if the parents were more stressed or if their child was more severely impaired, or was a girl, or had a higher EDS. However, these factors were correlated. Thus, in multivariable models only walking ability and EDS remained significantly associated with pain. We found no evidence of interaction between walking ability and EDS. Sensitivity analyses yielded similar results.

In order to understand better these differences between self-reported and parent-reported pain, we restricted analysis of parent-reported pain to young people who could self-report. Results in this sub-sample were similar to those in the complete sample: in particular, the relationship between parent-reported pain and walking ability remained significant.

Comparison of parents' reports of their child's pain and young people's reports of pain

The relationship between parent- and young person-reports of pain, (as captured by the instrument described in Measures

(1) in the Methods section), is shown in table 4. If parent- and self-report had agreed completely, the percentages in bold would be 100%. Although parent- and self-reported pain were significantly correlated (Spearman rank correlation=0.45, $p < 0.0001$), parents tended to overestimate their child's pain if self-reported pain was infrequent or mild and underestimate it if the self-reported pain was frequent or severe.

DISCUSSION

Main findings

About three quarters of young people aged 13 to 17 years with CP had pain in the previous week according to both self- and parent-reports. Forty per cent experienced pain in the legs and over one quarter experienced pain in the head, stomach or back, pain on moving or short unexpected pains. Almost half of the young people who had received physiotherapy had experienced pain during therapy; about a quarter of those who received botulinum injections experienced pain. Girls generally reported more pain than boys. More pain was associated with more emotional difficulties according to both the self- and parent-reports. Parents tended to report that their child had more pain if the child was more severely impaired; this trend was much less evident for self-reported pain.

Strengths and weaknesses

Participants were representative of all young people with CP across the range of severity of impairment, as they were sampled from nine geographic areas, eight of which had population based registers. Analyses of trends and associations did not dichotomise pain; they used ordinal regression based on information in all six categories of pain. Estimation of prevalence required dichotomisation and we chose the cut-point as no pain/any pain in order to allow comparison with other papers

Table 4 Relationship between self-reported and parent-reported pain in previous week (n=421)

| Self-report of pain by young person | | Parent-report of their child's pain | | | | | | | | | | | | | |
|-------------------------------------|----|-------------------------------------|----|---------------|----|-------------|----|--------------|---|------------|----|-------------|-----|--------|--|
| | | None of the time | | Once or twice | | A few times | | Fairly often | | Very often | | Every day | | Total | |
| Frequency of pain | | | | | | | | | | | | | | | |
| None of the time | 66 | (50%) | 33 | (25%) | 20 | (15%) | 4 | (3%) | 3 | (2%) | 5 | (4%) | 131 | (100%) | |
| Once or twice | 36 | (26%) | 49 | (35%) | 33 | (24%) | 9 | (6%) | 5 | (4%) | 8 | (6%) | 140 | (100%) | |
| A few times | 10 | (13%) | 24 | (31%) | 25 | (32%) | 5 | (6%) | 7 | (9%) | 7 | (9%) | 78 | (100%) | |
| Fairly often | 3 | (11%) | 7 | (26%) | 6 | (22%) | 5 | (19%) | 2 | (7%) | 4 | (15%) | 27 | (100%) | |
| Very often | 2 | (13%) | 0 | (0%) | 4 | (25%) | 5 | (31%) | 3 | (19%) | 2 | (13%) | 16 | (100%) | |
| Every day | 0 | (0%) | 2 | (7%) | 7 | (24%) | 4 | (14%) | 5 | (17%) | 11 | (38%) | 29 | (100%) | |
| | | None | | Very mild | | Mild | | Moderate | | Severe | | Very severe | | Total | |
| Severity of pain | | | | | | | | | | | | | | | |
| None | 66 | (52%) | 31 | (24%) | 15 | (12%) | 13 | (10%) | 3 | (2%) | 0 | (0%) | 128 | (100%) | |
| Very mild | 24 | (26%) | 23 | (24%) | 28 | (30%) | 17 | (18%) | 2 | (2%) | 0 | (0%) | 94 | (100%) | |
| Mild | 14 | (15%) | 18 | (19%) | 28 | (30%) | 24 | (26%) | 9 | (10%) | 1 | (1%) | 94 | (100%) | |
| Moderate | 9 | (15%) | 8 | (13%) | 13 | (21%) | 23 | (37%) | 8 | (13%) | 1 | (2%) | 62 | (100%) | |
| Severe | 3 | (12%) | 2 | (8%) | 4 | (15%) | 9 | (35%) | 7 | (27%) | 1 | (4%) | 26 | (100%) | |
| Very severe | 1 | (6%) | 2 | (12%) | 4 | (24%) | 4 | (24%) | 2 | (12%) | 4 | (24%) | 17 | (100%) | |

If parent- and self-report had agreed completely, the percentages in bold would be 100%.

on CP^{1 2 22} and papers on pain in the general population.^{23 24} Although non-response by families was 37% in SPARCLE1, and 27% of those in SPARCLE1 dropped out in SPARCLE2, sensitivity analyses adjusting for factors associated with non-response yielded similar results.^{8 9 21} The weights, which reflected the sampling design, allowed extrapolation from the reports of pain in our sample to an estimate of the population prevalence of pain in young people with CP.

Comparison with other studies

A study of parent-reported pain in the previous week in the general population of 7 to 17-year-olds found 15% had headaches, 8% abdominal pain and 5% back pain,²³ much lower than our rates. A study of 10 to 18-year-olds found the prevalence of self-reported pain in the previous week to be 34%,²⁴ lower than the 74% we report. Thus, young people with CP generally experience more pain than those without, and this has clear clinical implications.

Studies in young people of a similar age with CP report lower prevalence of pain than we do—for example, 56% in 11 to 18-year-olds¹ and 62% in 8 to 18-year-olds²—even though they asked about pain over the previous month rather than the previous week. However, our study was larger, more representative, distinguished self- and parent-reported pain and asked about pain related to healthcare procedures. One study¹ found, as we did, that a higher proportion of girls reported pain (64% and 50%, respectively). A similar gender difference is also found in the general population,²² and may be due to neurophysiological differences.²⁵ The association of pain with emotional difficulties in 13 to 17-year-olds is consistent with findings in young people without disability; for example, young people with severe chronic pain reported high levels of anxiety and depression.²⁶ If the association is causal, then the direction is unclear and may indeed be in both directions.

Between childhood and adolescence, the prevalence of self-reported pain increased from 60% (95% CI 54% to 65%)⁴ to 74% (95% CI 69% to 79%).

Implications

The prevalence of pain in young people with CP is high. This is important, not only because of the unpleasantness of pain but also because of the strong association of pain in CP with lower subjective well-being and reduced participation.^{5 6} Given that pain is so prevalent among young people with CP, strategies to reduce it must be either absent or inadequate.

CP is often accompanied by painful secondary musculoskeletal problems such as hip subluxation or dislocation, spinal scoliosis and muscle contractures in the upper and lower limbs. Furthermore, common clinical interventions, such as gastrostomy tubes, botulinum injections, assistive devices and physiotherapy, may cause pain; we found that almost half of those receiving some form of therapy reported experiencing pain during therapy. It is difficult to justify therapy that causes pain unless good evidence indicates that it improves some aspect of a young person's life—so clinicians should carefully consider the efficacy of therapies that have the potential to cause pain. A study of children with CP²⁷ found that assisted stretching was the daily activity most frequently identified as painful. This is especially worrying in the light of recent reviews of the effectiveness of passive stretching in people with CP,^{28–30} which conclude that stretching does not produce clinically important change in contractures, function or disability. A further review found little evidence for any benefit of postural management but evidence of disadvantages, including pain.³¹ We recommend that adverse effects and benefits of treatment are recorded and assessed to inform effective use of such treatments.

At consultations, clinicians should ask parents and young people directly about their pain, and should develop pain management plans incorporating preventive and responsive elements. For instance botulinum toxin may help hip pain,³² intrathecal baclofen may help painful spasms,³³ pain during physiotherapy can be minimised^{34 35} and cognitive behavioural therapy may help coping with pain. Emotional health and pain are associated, so clinicians should assess if emotional factors could be exacerbating pain.

As most people with CP live well into adulthood, our findings are also important for adult care.

Future research

Future research should focus on identifying the type of pain experienced and the elements of therapy that appear to cause it. In particular, these studies should assess how much stretching and mobilisation is required to maintain function or range of motion and whether can this be achieved without significant pain. In order to do this, more studies will need to be undertaken in the clinical setting.

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Contributors AC is the corresponding author and guarantor of the article; participated in the planning of the study, coordinated the study and took overall responsibility for the delivery of the work; participated in writing the paper and approved the final version; had full access to all the data in the study and had final responsibility for the decision to submit for publication. KNP was responsible for the day to day administration of the study and data collection in one centre, participated in maintaining the quality of the data, attended workshops, and drafted the paper. HOD performed all statistical analysis, participated in maintaining the quality of the data and in writing the paper; saw and approved the final version. CA participated in the planning of this study, was responsible for data collection in one centre, attended workshops and planning analysis and saw and approved the final version. AL participated in the planning of this study, was responsible for data collection in one centre, attended workshops and planning analysis and saw and approved the final version.

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Data sharing statement The data from this paper and the associated dataset can be examined in more detail on request from the corresponding author, Allan Colver. The data from the wider SPARCLE study will be available for secondary analyses once the the SPARCLE collaborators have completed their planned analyses; this will be in about 18 months.

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Pain in young people aged 13 to 17 years with cerebral palsy: cross-sectional, multicentre European study

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