Recurrent pain in adolescents with cerebral palsy: a longitudinal population-based study

SELMA MUJEZINOVIĆ LARSEN[1,2] 🆔 | TERJE TERJESEN[2,3] | REIDUN B JAHNSEN[1,4] | KJERSTI RAMSTAD[1]

1 Division of Paediatric and Adolescent Medicine, Oslo University Hospital, Oslo; 2 Faculty of Medicine, Institute of Clinical Medicine, University of Oslo, Oslo; 3 Division of Orthopaedic Surgery, Rikshospitalet, Oslo University Hospital, Oslo; 4 Faculty of Medicine, Research Centre of Habilitation and Rehabilitation Models and Services, Institute of Health and Society, University of Oslo, Oslo, Norway.

Correspondence to Selma Mujezinović Larsen at Oslo Universitetssykehus HF, Rikshospitalet, Barne-og ungdomsklinikken, Postboks 4950 Nydalen, N-0424 Oslo, Norway. E-mail: uxsemu@ous-hf.no

AIM To investigate the pain characteristics, pain interference with activities of daily living, and use of analgesics in adolescents with cerebral palsy (CP) and compare the results with previous findings.

METHOD Sixty-seven adolescents (median age 14y 4mo, range 12y 2mo–17y, 28 females, 39 males) classified in Gross Motor Function Classification System (GMFCS) levels III to V, who participated in a CP surveillance programme, were assessed on pain measures twice, 5 years apart. Primary caregivers marked recurrent pain sites and graded pain interference with activities of daily living and sleep. Information on pain severity was obtained through two questions from the Child Health Questionnaire (CHQ) and were transformed into a pain score scaled from 0 to 100, where 100 represented no pain. The use of short-acting analgesics was recorded.

RESULTS Over 5 years, the prevalence of recurrent pain, number of pain sites, pain intensity, and pain frequency all increased significantly. The most frequent pain sites were the hip/thigh in GMFCS level V and knee in GMFCS level III. The median CHQ pain score decreased from 60 to 40 ($p<0.001$). Pain interference with activities of daily living increased ($p=0.011$) but not for sleep. Twenty-eight of 54 participants with moderate or severe pain (CHQ pain score ≤60) received no short-acting analgesics.

INTERPRETATION In adolescents with CP, pain increased over 5 years despite follow-up in a surveillance programme. For enhanced management of pain, we propose that an algorithm on pain should be included in surveillance programmes.

Parents and medical professionals consider pain a highly important target for interventions in adolescents with cerebral palsy (CP).[1] A systematic review reported pain prevalence up to 75%.[2] Prevalence was higher in adolescents than in children and individuals with greater motor impairment.[3,4] Furthermore, individuals with more severe CP tended to have more intense and more frequent pain.[3] A recent cross-sectional, register-based study revealed that pain prevalence at different sites varied for different levels of motor impairment, with more hip/thigh pain in individuals with greater motor impairment, more knee pain in individuals needing walking aids, and more lower leg/foot pain in those with less motor impairment.[4] Importantly, pain influenced societal participation and quality of life negatively.[5–7] According to the National Institute for Health and Care Excellence (NICE) guidelines on CP, pain should be addressed at each clinical encounter.[8]

Longitudinal studies on pain characteristics and interference with activities of daily living in the paediatric population with CP would be useful for patient education, pain management, and improvement of surveillance programmes; however, such studies are scarce.

With regard to pain management, the use of short-acting analgesics varies from one in three to one in four patients.[7,9] In both studies, the proportion of the population with pain was greater than the proportion receiving analgesics, indicating that the full potential of analgesics might not be fully exploited. In line with this, a retrospective study confirmed that pain reported repeatedly in a CP surveillance programme was largely neglected in corresponding medical records.[10] This indicates that we need to reconsider both how we assess pain in CP surveillance programmes and bridge the assessments into pain management.

The aims of the present study were to investigate pain characteristics, pain interference with activities of daily living, and the use of short-acting analgesics in a cohort of adolescents participating in a CP surveillance programme and compare the results with findings reported 5 years earlier.

METHOD

The study had both a cross-sectional and longitudinal design. All 136 eligible adolescents, born between 2002 and 2006, living in south-eastern Norway, and enrolled in the Norwegian Quality and Surveillance Registry for Cerebral Palsy,[11] with bilateral CP and in Gross Motor Function Classification System (GMFCS) levels III to V[12] were invited to participate.[13] Data on CP type according to the Surveillance of Cerebral Palsy in Europe,[14] communication function according to the Communication Function Classification System,[15] and gross motor function according to the GMFCS[12] were retrieved from the Norwegian Quality and Surveillance Registry for Cerebral Palsy.[11]

The study was approved by the Regional Ethics Committee, REC South-East (no. 2012/2258 REK). Written informed consent was obtained for 77 adolescents (57%) in 2013 to 2014.[13] Six participants were lost to follow-up 5 years later. Thus, 71 participants received a postal invitation to the second data collection; of these, 67 (94%) participated.

Pain assessment

Data were collected through a questionnaire sent by surface mail to primary caregivers and a telephone interview. The questionnaire consisted of selected questions from the Brief Pain Inventory (BPI), Norwegian version[16] and Child Health Questionnaire (CHQ), Norwegian version.[17] The CHQ has been validated for CP;[18] the reliability of proxy reporting on pain interference in severe CP according to the BPI has been found to be satisfactory.[19]

Pain occurring for at least 4 weeks or more was defined as 'recurrent pain' and further noted as 'pain'. Pain sites with recurrent pain were marked on the BPI body outline.

Pain severity according to the CHQ was recorded for the most severe pain site (selected by the respondent). The two CHQ questions were (1) 'During the past 4 weeks, how much bodily pain or discomfort has your child had?' with the response alternatives 'none, very mild, mild, moderate, severe, and very severe', and (2) 'During the past 4 weeks, how often has your child had bodily pain or discomfort?' with the response alternatives 'none of the time, once or twice, a few times, fairly often, very often, and every day or almost every day', and were given scores from 1 to 6 respectively. These scores were transformed by an algorithm into a CHQ pain score scaled from 0 to 100, where 100 represented no pain.[20] After careful consideration and with the aim of defining a pain scoring system feasible for recommendations in a CP surveillance protocol, we categorized CHQ pain scores as 0 to 30 (severe pain), 40 to 60 (moderate pain), and 70 to 90 (mild pain). A change in CHQ pain score of 20 and less was considered as no change in pain, while a change of 30 or more was regarded as less or more pain.

The BPI questions on pain interference with activities of daily living and sleep were: 'On a scale from 0 to 10 (10=total influence), which value best describes how much pain influenced your child's 'activities of daily living' and

What this paper adds
- Pain prevalence and the number of pain sites increased over a 5-year period in adolescents with cerebral palsy.
- Pain intensity, frequency, and pain interference with activities of daily living increased, whereas interference with sleep was unchanged.

'sleep' respectively?' The time span was modified from 2 to 4 weeks to correspond with the CHQ.

The telephone interview started with the definition of recurrent pain and consisted of the following questions with an open response: Did your child have recurrent pain in the last 4 weeks? What are the pain sites? What relieves the pain? What increases the pain? Has your child received any medication to relieve pain (such as paracetamol, ibuprofen, or naproxen) over the past 4 weeks? Use of intrathecal baclofen (ITB) was also recorded.

Statistical analysis

SPSS v27 (IBM Corp., Armonk, NY, USA) was used for the statistical analysis. Data were presented either as frequency, percentage, proportion, or median with range. Correlation between variables was explored by calculating Spearman's correlation coefficient, r_s (significant when $p<0.05$ and $r_s>0.30$). Non-parametric statistics (Mann–Whitney U test) were applied for ordinal variables and skewed continuous variables. In linear regression, variables were included in the multivariate linear regression analyses if $p<0.1$ in the univariate analyses. Normality of residuals was satisfied. In the longitudinal analyses, proportions were analysed with McNemar's test, continuous variables with a paired samples t-test, and ordinal variables with a Wilcoxon signed-rank test. All tests were two-sided. Differences were significant if $p<0.05$.

Three participants (4.4%) had missing values on pain interference at the first data collection point. Imputation was not performed since this would most likely not influence the statistical analyses.

RESULTS

The characteristics of the participants are shown in Table 1. The number of participants with pain increased from 45 to 62 over the 5-year period ($p<0.001$) and the median number of pain sites in each participant increased from one (range 0–6) to three (range 0–13; $p<0.001$).

Pain prevalence increased at all sites and the increase was statistically significant in the neck and knee (Table 2). Pain prevalence increased across all GMFCS levels and the increase was statistically significant in GMFCS level III. The hip/thigh was the most common pain site in GMFCS level V, while the hip/thigh and lower leg/foot were the most common sites in GMFCS level IV and the knee in GMFCS level III. The prevalence of abdominal pain was highest in GMFCS level V.

There was a significant correlation between pain intensity and pain frequency ($r_s=0.494$, $p<0.001$). Both pain intensity and frequency increased during the 5-year period (both $p<0.001$). The median CHQ pain score decreased from 60 to 40 ($p<0.001$). Decrease across GMFCS levels

Table 1: Characteristics of the 67 participants

Characteristic	
Age, y:mo, median (range)	14:4 (12:2–17:0)
Sex	
Female	28 (42)
Male	39 (58)
Predominant movement disorder	
Spastic	53 (79)
Dyskinetic	14 (21)
Communication	
CFCS level I	7 (10)
CFCS level II	10 (15)
CFCS level III	2 (3)
CFCS level IV	13 (19)
CFCS level V	21 (31)
Unknown	14 (21)
Ambulation	
GMFCS level III	15 (22)
GMFCS level IV	17 (25)
GMFCS level V	35 (52)
Intrathecal baclofen therapy[a]	15 (22)

Data are n (%) unless otherwise stated. [a]Three of 17 participants in GMFCS level IV and 12 of 35 participants in GMFCS level V had intrathecal baclofen therapy. CFCS, Communication Function Classification System; GMFCS, Gross Motor Function Classification System.

Table 2: Longitudinal analysis on the prevalence of recurrent pain sites in 67 adolescents with cerebral palsy 5 years apart

Pain site	Present	Previous	p
Pain at any site	62 (93)	45 (67)	<0.001
GMFCS level III	12 (80)	6 (40)	0.031
GMFCS level IV	16 (94)	11 (65)	0.063
GMFCS level V	33 (94)	28 (80)	0.180
Neck pain	11(16)	1 (1)	0.006
GMFCS level III	1 (7)	0 (0)	0.334
GMFCS level IV	2 (12)	1 (6)	0.579
GMFCS level V	8 (23)	0 (0)	0.003
Back pain	12 (18)	6 (9)	0.109
GMFCS level III	3 (20)	1 (7)	0.164
GMFCS level IV	2 (12)	3 (18)	0.332
GMFCS level V	7 (20)	2 (6)	0.058
Upper-limb pain	18 (27)	10 (15)	0.152
GMFCS level III	3 (20)	1 (7)	0.334
GMFCS level IV	3 (18)	0 (0)	0.083
GMFCS level V	12 (34)	9 (26)	0.475
Hip/thigh pain	34 (51)	24 (36)	0.076
GMFCS level III	4 (27)	3 (20)	0.670
GMFCS level IV	8 (47)	6 (35)	0.431
GMFCS level V	22 (63)	15 (43)	0.070
Knee pain	31 (46)	18 (27)	0.035
GMFCS level III	9 (60)	1 (7)	0.006
GMFCS level IV	7 (41)	7 (41)	1.000
GMFCS level V	14 (40)	10 (29)	0.481
Lower leg/foot pain	21 (31)	18 (27)	0.678
GMFCS level III	6 (40)	3 (20)	0.082
GMFCS level IV	8 (47)	5 (29)	0.188
GMFCS level V	7 (20)	10 (29)	0.447
Abdominal pain	13 (19)	9 (13)	0.344
GMFCS level III	0 (0)	0 (0)	–
GMFCS level IV	2 (12)	2 (12)	1.000
GMFCS level V	11 (31)	7 (20)	0.160
Headache[a]	4 (6)	3 (4)	1.000
Other pain sites[a]	10 (15)	4 (6)	–

Data are n (%) unless otherwise stated. [a]Analyses for GMFCS levels not performed due to small numbers. Pain in the teeth and skin or pain localized to the respiratory or genito-urinary system was merged and labelled as 'other pain sites'. A McNemar' test was used to compare proportions. GMFCS, Gross Motor Function Classification System.

was: GMFCS level III, 100 to 40 (p=0.002); GMFCS level IV, 60 to 50 (p=0.050); and GMFCS level V, 50 to 30 (p=0.007). There were 34 (51%) participants with more pain (lower CHQ score) than 5 years before, 27 (40%) with no change, and six (9%) with less pain. In the univariate analyses, lower age and GMFCS level V were possible predictors of low CHQ score (B=7.8, 95% confidence interval [CI]=3.6–12.1, p<0.001 and B=−16.6, 95% CI=−33.1 to −0.1, p=0.049 respectively), while sex and predominant movement disorder were not (B=−5.9, 95% CI=−19.4 to 7.7, p=0.392 and B=−3.9, 95% CI=−20.4 to 12.7, p=0.643 respectively). In the multivariate analysis, younger age was the only independent predictor of lower CHQ scores (B=7.1, 95% CI=2.7–11.4, p=0.002).

There was a positive correlation between pain interference with activities of daily living and sleep (r_s=0.762, p<0.001) and negative correlations between CHQ pain score and both interference with activities of daily living and sleep (r_s=−0.521, p<0.001 and r_s=−0.370, p=0.002 respectively). Participants classified in GMFCS level III had significantly lower interference with activities of daily living (GMFCS level III vs GMFCS level IV p=0.043 and GMFCS III vs GMFCS level V p=0.001) and sleep (GMFCS level III vs GMFCS level IV p=0.009 and GMFCS level III vs GMFCS level V p<0.001) than participants in GMFCS levels IV and V. There were no significant differences between GMFCS levels IV and V regarding pain interference with activities of daily living and sleep (p=0.235 and p=0.050 respectively). Median pain interference with activities of daily living increased over the 5-year period from 1.5 (range 0–10) to 3.0 (range 0–10) (p=0.011), while median pain interference with sleep

was 1.0 (range 0–10) at both data collection time points (p=0.767) in 64 participants.

The relationship between CHQ pain score and the use of analgesics is shown in Table 3. Twenty-six participants received analgesics and 41 did not. Their median CHQ pain score was 20 (range 0–60) and 50 (range 10–100) respectively (p<0.001). Regarding changes in pain, 18 of 34 participants with more pain did not receive analgesics and neither did 8 of the 12 participants with a pronounced pain increase (CHQ score decrease ≥60). There was a positive correlation between the use of analgesics and pain interference with activities of daily living and sleep (r_s=0.682, p<0.001 and r_s=0.415, p<0.001 respectively).

The median CHQ pain score was 30 (range 10–100) in the 15 participants receiving ITB. Ten of 15 participants receiving ITB received analgesics and five did not. Their median CHQ pain score was 20 (range 10–60) and 70 (range 30–100) respectively.

Longitudinal data on pain characteristics and interference are available in Figure S1 and Table S1 (online

Table 3: Caregiver-administered short-acting analgesics during the last 4 weeks in relation to pain severity at the two data collection points

Pain score	Present		Previous	
	Medication n=26	No medication n=41	Medication n=12	No medication n=55
CHQ 100	0	8	1	20
CHQ 70–90	0	5	1	10
CHQ 40–60	11	16	7	20
CHQ 0–30	15	12	3	5

Medication consisted of short-acting analgesics (paracetamol/ibuprofen/naproxen). CHQ, Child Health Questionnaire.

supporting information). Caregiver experiences on factors that increased and relieved pain are shown in Table S2 (online supporting information).

Forty-five caregivers gave a written response to the BPI and CHQ and were interviewed, while 22 caregivers responded to the BPI and CHQ during the telephone interview. Responders (87% parents, 85% female) were the same 61 caregivers as 5 years earlier as well as six new primary caregivers in the home care facility. The mean time between the two data collection points was 5 years 1 month (median 5y 2mo, range 3y 8mo–5y 11mo).

DISCUSSION
The main findings of this study were that the prevalence of recurrent pain was high in adolescents with CP and that pain prevalence, the number of pain sites, pain severity, and pain interference with activities of daily living all increased over a period of 5 years despite follow-up in a CP surveillance programme.

Pain prevalence was higher (93%) than in previous studies (37–77%).[2,3] One reason could be that we recorded pain regardless of pain severity or level of pain interference. Also, our population was restricted to ages 12 to 17 years and GMFCS levels III to V, a group with expected high pain prevalence.[4] The increase in pain prevalence was statistically significant only in GMFCS level III, which could be attributed to weight gain, extensive physical strain, and increasing contractures.[21]

Pain prevalence at each site was higher than reported by Eriksson et al.,[4] possibly due to a narrower age range in our study population. In line with previous studies, the most frequent pain site was the lower limbs.[3,4] A trend of most frequent pain in the abdomen and hip/thigh in GMFCS level V and most frequent knee pain in GMFCS level III was supported.[4] Furthermore, increasing frequency of neck pain in GMFCS level V and knee pain in GMFCS level III was reported. Our data on neck pain must be taken with caution since this group consisted of only 11 participants. Nonetheless, positional factors such as lack of adjustment of the wheelchair and prolonged sitting or lying without support are potential causes. An increase in knee pain could be caused by increased crouch gait and lack of correct adjustment or omitted use of
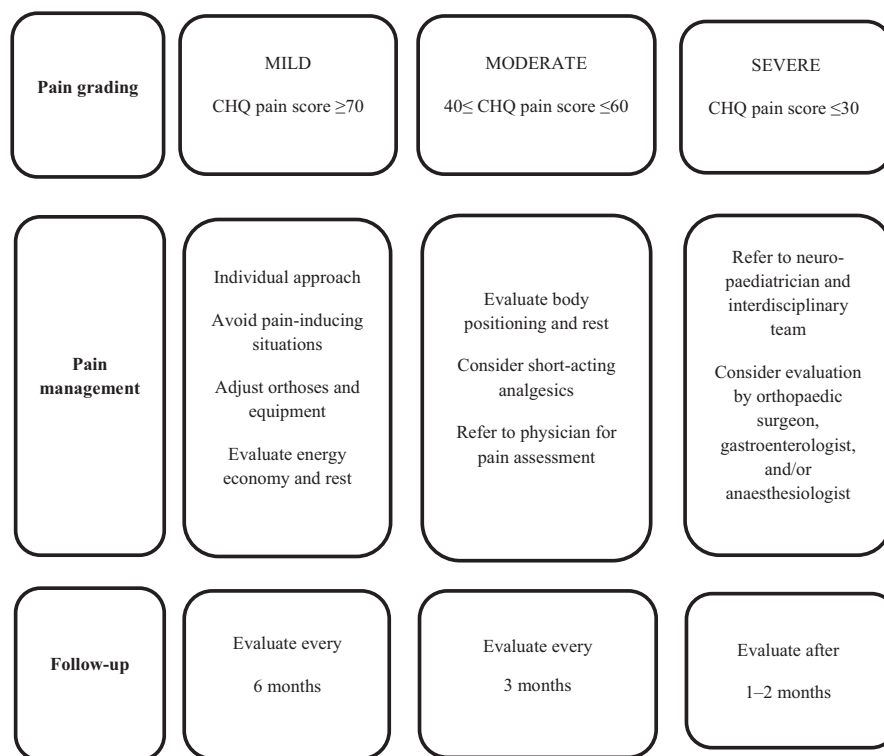
orthoses, which is common in adolescents in GMFCS level III.[22]

Our participants had moderate-to-severe pain (60%) and daily pain (40%) more often than previously reported by Parkinson et al.[3] (37% and 11% respectively), which is probably explained by the inclusion of only GMFCS levels III to V in the present study. The finding of younger age as a predictor of more severe pain within the 12 to 17 year age group is not in agreement with the study by Eriksson et al.[4] One reason could be the narrow age range. We found no significant difference in pain severity between participants with spastic and dyskinetic bilateral CP.

Pain interfered with activities of daily living, thus adding to the knowledge base that pain in CP has a negative impact on daily life.[4,7,23] The finding of higher pain interference with activities of daily living in GMFCS levels IV and V was in line with the study by Christensen et al.[24]

Longitudinal studies are useful to evaluate the natural history of pain and the effects of treatments and interventions. Our study had an observation time of 5 years, which ensured a comparison between childhood and adolescence in the same individuals. Two longitudinal studies previously reported changes in pain in a paediatric population with CP.[24,25] These studies reported no significant change in mean pain scores. In contrast, we found a more adverse pain development, with an increase in pain and pain interference with activities of daily living over 5 years. Comparing the results is difficult because of differences in study population (population- vs hospital-based), age (mean=14y 7mo vs 8y 8mo vs 8y 6mo), age range (12y 2mo–17y vs 3–16y vs 3–18y), time span between data collection points (mean=5y 1mo vs 2y 4mo vs 1y), and inclusion of GMFCS levels III to V only versus including all GMFCS levels.[24,25]

Factors that aggravated pain (Table S2) were in line with a previous report.[3] The most prevalent factor was staying in the same position over time. This information should be discussed with primary caregivers to secure 24/7 positioning strategies. Passive muscular stretching both relieved and increased pain. Other actions that most often relieved pain were rest, change of position, and use of analgesics. In line with the study by Tedroff et al.,[9] who reported a positive correlation between the frequency of use of analgesics and pain interference, we found positive correlations between the use of analgesics and both pain interference with activities of daily living and sleep. Although the number of participants receiving analgesics increased over 5 years, 28 out of 54 participants with moderate or severe pain did not receive analgesics. This suggests that the pain-relieving potential of analgesics was not fully exploited. Nonetheless, the use of analgesics should be based on individual preference and potential side effects as well as pain severity and interference. We have not been able to find studies on pain and the use of analgesics in children with CP receiving ITB. The finding that most had moderate or severe pain and received analgesics indicates the need for close follow-up of pain even if ITB is used.

Pain grading	MILD CHQ pain score ≥70	MODERATE 40≤ CHQ pain score ≤60	SEVERE CHQ pain score ≤30
Pain management	Individual approach Avoid pain-inducing situations Adjust orthoses and equipment Evaluate energy economy and rest	Evaluate body positioning and rest Consider short-acting analgesics Refer to physician for pain assessment	Refer to neuro-paediatrician and interdisciplinary team Consider evaluation by orthopaedic surgeon, gastroenterologist, and/or anaesthesiologist
Follow-up	Evaluate every 6 months	Evaluate every 3 months	Evaluate after 1–2 months

Figure 1: Algorithm on the grading, management, and follow-up of recurrent pain. The Child Health Questionnaire (CHQ) pain score was calculated according to the CHQ manual. The pain management suggested for mild pain is relevant for moderate and severe pain; the pain management suggested for moderate pain is relevant for severe pain.

Data from Sweden show that the prevalence of hip dislocation declined after the introduction of a surveillance programme that included an algorithm on hip management.[26] In contrast, the high prevalence of recurrent pain suggests that our CP surveillance programme was not helpful in initiating adequate pain management. This is in line with the study by Westbom et al.[10] After this study, the Nordic CP surveillance protocols were revised and questions on pain intensity, frequency, and interference with activities of daily living and sleep have since been included.[11] Also, the Swedish CP surveillance protocol now includes questions on pain intensity for each pain site. We support these inclusions and suggest that information on laterality of pain in the limbs and pain duration should be included. We propose that an algorithm on pain assessment and management based on the CHQ pain score should be included in the surveillance protocol (Fig. 1). One could consider differentiation in GMFCS levels in the algorithm, such as more detailed assessment of hip pain in GMFCS level V, since hip pain might be an indicator for surgical treatment of hip subluxation if the migration percentage is equal to or greater than 40%.[27] Physical assessment by a physician should be included in cases of moderate and severe pain. The local multidisciplinary team should consider all causes of pain and all available treatment alternatives and outline an action plan for each pain site. The proposed

algorithm should be adjusted to the local health care system to be feasible.

This study has several limitations. First, data were based on proxy reports of pain; by definition, pain is subjective and should be self-reported whenever possible. Most of our population was in Communication Function Classification System levels IV and V; therefore, proxy reporting had to be applied to include the whole sample. Furthermore, a recent study found no significant differences in self- versus proxy-reported pain in a CP surveillance programme.[28] Second, we did not ask about the duration of pain episodes. This information could have contributed to the understanding of why some participants with moderate and severe pain did not receive analgesics. Third, the questionnaires sent out in paper form were answered during a telephone interview by one-third of primary caregivers. However, there were no significant differences in the number of pain sites or CHQ pain scores with regard to the response form. Finally, interviews were performed by a different researcher than 5 years earlier, which could challenge the reliability of longitudinal comparison.

The study has several strengths. First, it is population-based and there were no significant differences between participants and non-responders.[13] This ensured generalizability of the data. Second, the response rate at the second data collection point was high, probably because the

method included a telephone interview. Furthermore, most responders were the same person at the two data collection points, which strengthens the reliability of the comparison between the two time points.

In conclusion, pain is a considerable problem in adolescents with CP. We propose extended pain assessment and an algorithm on pain management to be included in CP surveillance programmes with the aim to bridge the gap between programmes, guidelines, and pain management.

ACKNOWLEDGEMENTS

CONFLICT OF INTEREST

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

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

SUPPORTING INFORMATION

The following additional material may be found online:

Figure S1: Longitudinal data on the number of pain sites, CHQ pain score, pain interference with daily activities and sleep, pain intensity, and pain frequency in 67 participants with CP 5 years apart.

Table S1: Longitudinal data on pain characteristics in adolescents with CP collected 5 years apart

Table S2: Proxy-reported factors which relieved and increased pain (open response)

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