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Pain in children with dyskinetic and mixed dyskinetic/spastic cerebral palsy

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ABBREVIATIONS

FPS-R	Faces Pain Scale-Revised
HUI-3	Health Utilities Index-3
PPP	Paediatric Pain Profile

[abstract]

AIM To evaluate pain prevalence and characteristics in children and adolescents with predominant dyskinetic and mixed (dyskinetic/spastic) cerebral palsy (CP) motor types.

METHOD Seventy-five participants with a diagnosis of CP and confirmed dyskinetic or mixed (dyskinetic/spastic) motor type took part in a multisite cross-sectional study. The primary outcome was carer-reported pain prevalence (preceding 2wks) measured using the Health Utilities Index-3. Secondary outcomes were chronicity, intensity, body locations, quality of life, and activity impact.

RESULTS Mean participant age was 10 years 11 months (SD 4y 2mo, range 5–18y). There were 44 males and 31 females and 37 (49%) had predominant dyskinetic CP. Pain was prevalent in 85% and it was chronic in 77% of participants. Fifty-two per cent experienced moderate-to-high carer-reported pain intensity, which was significantly associated with predominant dyskinetic motor types (p=0.008). Pain occurred at multiple body locations (5 out of 21), with significantly increased numbers of locations at higher Gross Motor Function Classification System levels (p=0.02). Face, jaw, and temple pain was significantly associated with predominant dyskinetic motor types (p=0.005). Poorer carer proxy-reported quality of life was detected in those with chronic pain compared to those without (p=0.03); however, chronic pain did not affect quality of life for self-reporting participants.

INTERPRETATION Pain was highly prevalent in children and adolescents with predominant dyskinetic and mixed (dyskinetic/spastic) motor types, highlighting a population in need of lifespan pain management.

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What this paper adds

- Chronic pain prevalence in children and adolescents with predominant dyskinetic and mixed (dyskinetic/spastic) motor types is high.
- Pain occurs across multiple body locations in predominant dyskinetic and mixed (dyskinetic/spastic) motor types.
- Less recognized locations of pain include the face, jaw, and temple for predominant dyskinetic motor types.

[main]

Cerebral palsy (CP) is a heterogeneous disorder of posture and movement classified according to its predominant neuromotor disability into spastic, dyskinetic, or ataxic motor types.[1,2] Children with predominant dyskinetic motor types account for 7% of the Australian population with CP, often presenting with high levels of physical disability due to total body involvement, marked functional limitations, and frequent medical comorbidities including pain.[3–6] Pain is a complex sensory experience influenced by a range of cognitive, affective, and behavioural mechanisms and measured according to biopsychosocial pain models.[7,8] It can be described as

being acute, short-lasting, or chronic (lasting >3mo).[7] In broader CP populations, pain prevalence is reported to vary from 14% to 76% and is more likely in adolescents compared to younger children, females, and those with severe motor impairments.[9] However, the influence of pain on children and adolescents with dyskinetic CP is underexplored within current CP pain research, with a need to build further knowledge in this area to inform management.

Dyskinetic CP is characterized by involuntary, uncontrolled, and recurring movements with fluctuating muscle tone; it is further differentiated as dystonia and choreoathetosis.[1,2] Dystonia and choreoathetosis often present simultaneously; however, dystonia is more dominant and commonly classified.[10] There is emerging recognition that some children present with both dystonia and/or choreoathetosis as well as spasticity; they are described as having a mixed motor type.[11] Both children with mixed and dyskinetic motor types have been reported to be at risk of pain, with a need to explore pain characteristics within both of these important subpopulations.[12]

Pain management is complicated by multifactorial pain sources and mechanisms that place children with predominant dyskinetic and mixed (dyskinetic/spastic) CP motor types at risk of under detected and undertreated pain.[13,14] As children grow, pain may be caused by non-invasive management (e.g. physiotherapy, stretching), invasive management (e.g. surgery), general health conditions (e.g. headaches, period pain), and other CP-related comorbidities.[15] CP-related comorbidities are characteristically high in predominant dyskinetic motor types, including scoliosis, hip displacement, muscle contracture, and gastrointestinal dysfunction, as well as the movement disorder itself.[16–18] Dystonia causes pain due to the fluctuating and intermittent nature of muscle contractions resulting in abnormal and torsional body movement and postures.[19] Dystonic posturing may exacerbate pain due to suboptimal body alignment and skin irritation in supportive equipment. Over time, dystonia at extreme ranges may lead to repetitive joint stress, causing altered joint biomechanics and possible early-onset

osteoarthritis.[20] Unpredictable responses to orthopaedic management (i.e. soft tissue surgery) may further prolong pain due to longer recovery times and complications post-surgery.[2,21] Pain in dyskinetic populations may be further complicated by complex bidirectional interactions between pain and dystonia or choreoathetosis. In these interactions, pain sources may trigger exacerbations of dystonia or choreoathetosis, while dystonia or choreoathetosis may also cause pain in amongst other possible stimuli. These multifactorial contributors to pain presentations complicate pain diagnosis and treatment and are experienced within the context of developing children with complex neurodevelopmental disability. There is a need to learn more about the true impact of pain amongst this complex population of children with dystonia and/or choreoathetosis, to help distinguish their needs from the broader CP population.

Coordinated, evidenced-based, and multidisciplinary pain management is lacking in children with CP.[22,23] For those with dystonia or choreoathetosis, medical management predominates, typically focusing on treating the underlying pain sources with or without pain-relieving medications, as well as the use of tone-alleviating interventions to target alterations in muscle tone and movement disorder.[23,24] However, little is known regarding the pain characteristics of children with CP and dystonia or choreoathetosis to help guide treatment. Therefore, the primary aim of this study was to evaluate pain prevalence in children and adolescents with predominant dyskinetic and mixed (dyskinetic/spastic) motor types. The secondary aim was to evaluate pain characteristics including chronicity, impact on activity, intensity, body locations, and quality of life.

METHOD

This prospective, multicentre, cross-sectional study was undertaken at the Royal Children's Hospital Melbourne and Monash Children's Hospital, both Australian tertiary paediatric hospitals in the state of Victoria. These hospitals provide a range of specialist tertiary level

services, with the Royal Children's Hospital Melbourne also operating a complex movement disorder clinic. Ethical approval was obtained from the Royal Children's Hospital Melbourne, Monash University, and Monash Children's Hospital Ethics Committees (no. HREC/17/RCHM/359).

Participants and recruitment

Recruitment occurred from January 2018 to March 2019. A consecutive sample of children and adolescents was identified by clinicians who screened outpatient clinic lists and the electronic medical record for potential participants meeting the eligibility criteria. Participants were eligible if they had a confirmed diagnosis of CP, were aged between 5 and 18 years, and had a dyskinetic or mixed (dyskinetic/spastic) motor type. Exclusion criteria were major neurosurgical (i.e. deep brain stimulation, intrathecal baclofen implantation, selective dorsal rhizotomy) or orthopaedic procedures (i.e. bony or soft tissue surgery) within the preceding 6 weeks or if the primary carer could not speak English. Children were recruited from the Victorian Paediatric Rehabilitation Service orthopaedic, neurodevelopment, and disability clinics at the Royal Children's Hospital Melbourne and the Victorian Paediatric Rehabilitation Service clinic at Monash Children's Hospital.

Procedures

Participants were assessed by an experienced research physiotherapist (either CM or two others) trained in the study protocol. The presence of a dyskinetic or mixed motor type was confirmed using the Hypertonia Assessment Tool[25] and Cerebral Palsy Description Form of the Australian Cerebral Palsy Register.[26] Before the study started, assessors familiarized themselves with tool administration guidelines, watched a demonstration video, and had a practical session to ensure reliability. Written informed consent was subsequently completed for confirmed eligible participants and demographic and pain survey data were collected. The

demographics collected included age, sex, Gross Motor Function Classification System (GMFCS) level, Communication Function Classification System level, Manual Ability Classification System level, presence of intrathecal baclofen pump/deep brain stimulation, seizures, gastrostomy, previous bony hip or spinal surgery, and the Socio-Economic Index for Areas Index of Relative Social Disadvantage 2016.

Outcome measures

A range of carer- and self-report outcome measures were collected. Some children self-reported if cognitively able; if not, measures were carer-reported as stated.[27] The capacity of a child to self-report was decided through mutual discussion between the researcher and carer; this considered age, maturity, communication competence and style, cognitive ability, the type of pain tool, and the recall period for pain. These multifactorial considerations meant that the capacity to self-report could vary across measures. For participants reliant on augmentative and alternative communication to self-report, carers were used as communication partners where necessary to enhance communication; participants were also given regular rest periods to minimize physical fatigue while being assessed.[28] Where participants could self-report, a flexible approach to pain tool administration was adopted to accommodate for varying communication, reading ability, vision, functional level, and physical fatigue. Self-reporting participants could defer to their carers during an assessment if needed, with reporting categorized as combination where both carer- and self-reporting were used.

All research assessments commenced with the carers completing the Health Utilities Index-3 (HUI-3) over the preceding 2 weeks, the measure for the primary outcome of pain prevalence.[29] The HUI-3 classification system (1–5) embeds pain severity assessment into its impact on activity.[19,30] A score equal to or greater than 2 indicates pain prevalence, while a score equal to or greater than 3 indicates that pain impacts activity levels. Subsequently, the

researcher classified chronic pain according to the International Association for the Study of Pain definition asking about the presence of ongoing (constant or recurrent) pain for the last 3 months, self-reporting if able or carer-reported if unable.[7] Lay language was used for self-reporting participants; the researcher also clarified pain history and perceived source to ensure accurate classification. The remaining outcome measures were administered using a flexible approach for self-reporting participants or self-administered with a researcher present to answer questions for carer proxy-reported outcomes.

Body locations of pain over the preceding 2 weeks were measured using the Childhood Arthritis and Rheumatology Research Alliance Body Diagram, which was self-reported if able or carer-reported if unable.[31] The Childhood Arthritis and Rheumatology Research Alliance Body Diagram defines 21 pain regions and is a valid and reliable paediatric chronic pain measure.[32] For children or adolescents able to self-report with adequate upper-limb function, the participant marked the body regions directly. Otherwise, the researcher established pain presence and marked on their behalf.

Pain intensity over the preceding 2 weeks was reported by carers using the Paediatric Pain Profile (PPP). The PPP is a 20-item, observational, behavioural rating scale scored from 0 to 60, with a score of 14 or more indicative of moderate-to-severe pain.[33] The 'ongoing' PPP form was used to give a single pain score and the 'on a good day' PPP form was used to record the child or adolescent's best level of pain intensity. The PPP is validated in children with CP unable to self-report their pain.[33] For children or adolescents able to self-report pain, the Faces Pain Scale-Revised (FPS-R) was completed over the preceding 2 weeks.[34] The psychometric properties of the FPS-R have been evaluated in generic paediatric conditions but not in children with CP.[35]

Quality of life was measured using the Cerebral Palsy Quality of Life Questionnaire (self- or carer-reported).[36] The Cerebral Palsy Quality of Life Questionnaire uses a 0 to 100 scale for

each domain to give an overall quality of life score, with higher scores indicative of higher quality of life. It is valid and reliable for individuals with CP.[37]

Statistical analysis

Data were analysed using Stata v14.0 (StataCorp, College Station, TX, USA). For the study demographics, the mean and SD were used to describe age, while frequencies and proportions were used for the remaining demographics and FPS-R. Prevalence proportions and 95% confidence intervals (CIs) were used to describe HUI-3 distribution, chronicity, body locations, and level of pain intensity according to the PPP (score≥14). For binary outcomes (pain prevalence, pain presence at specific bodily locations, PPP>14), univariate logistic regression models were used and odds ratios (ORs) calculated. For the continuous outcome of the total PPP score, a univariate linear regression model was used, with regression coefficients reported. Within all univariate regression models, sex (male, female), subtype (mixed, dyskinetic), and age (5–12y, 13–18y) were included as individual predictors within models. The age ranges used to define children and adolescents were aligned with the Cerebral Palsy Quality of Life Questionnaire.[37] Before analysing the Cerebral Palsy Quality of Life Questionnaire scores, the normality of the distribution was evaluated using histograms and scatter plots to map scores. Since the distribution of self-reported Cerebral Palsy Quality of Life Questionnaire scores was not normally distributed, the Mann–Whitney U test was used to evaluate the between-group differences between self- and carer-reported total scores/domains, as well as the influence of chronic pain across the total self-reported sample. Since carer proxy-reported quality of life scores were normally distributed, an independent samples t-test was used to evaluate the influence of chronic pain across the total carer-reported sample. The independent samples t-test was also used to evaluate between-group differences in quality of life and the number of painful body locations (based on all 21 defined regions). Some body locations of the Childhood

Arthritis and Rheumatology Research Alliance Body Diagram were merged (leaving 14 regions) to minimize measurement error given the small sample and large number of defined regions.

RESULTS

Seventy-six of 128 children and adolescents approached agreed to take part. One child was excluded because dystonia or choreoathetosis was not present on screening, leaving 75 participants (59% response rate). Mean participant age was 10 years 11 months (SD 4y 2mo, range 5–18y), 44 were male, 31 were female, and 37 (49%) had predominant dyskinetic CP. Participant demographics are further described in Table 1. Across the cohort, high proportions of mixed motor types presented in the lower limbs, while both pure dystonic and mixed motor types were common in the upper limbs according to the Hypertonia Assessment Tool scores (Table 2).

Table 3 summarizes the outcome measures completed, reporting, and any missing data.

Pain prevalence and impact on activity

Pain prevalence (HUI-3≥2) over the preceding 2 weeks was 85% (95% CI 76–92%) across the cohort, occurring in 92% (95% CI 77– 97%) of dyskinetic and 79% (95% CI 63–89%) of mixed motor types. Chronic pain was present in 77% (95% CI 67–86%), occurring in 78% (95% CI 61–87%) of dyskinetic and 76% (95% CI 60–87%) of mixed motor types. Pain impacted activity levels (HUI-3≥3) in 49% (95% CI 38–60%) of participants, with no difference between those classified in higher (IV and V) and lower (I–III) GMFCS levels (mean difference [MD]=−0.03, 95% CI −0.29 to 0.24, p=0.85).

Body locations

Body locations of pain were most common in the lower limbs 69% (95% CI 57–79%) compared to the upper limbs 32% (95% CI 22–43%) (Fig. 1). On average, children had 5 out of 21 body locations of pain, with a significantly higher number at higher (IV and V) compared to lower (I–III) GMFCS levels (MD=−3.31, 95% CI −6.06 to 0.57, p=0.02) (Fig. 2). Participants with predominant dyskinetic motor types were more likely to experience face, jaw, and temple pain (OR=9.75, 95% CI 2.02–47.14, p=0.005) compared to those with mixed motor types.

Pain intensity

Carer-reported pain intensity was categorized as moderate-to-high according to the PPP (score≥14) in 40% (95% CI 29–52%) of participants on 'a good day' and 52% (95% CI 37–60%) in the preceding 2 weeks, and was significantly associated with predominant dyskinetic motor types (OR=3.69, 95% CI 1.40–9.70, p= 0.008). However, when analysing across the total PPP score, there was no significant difference between mixed and predominant dyskinetic motor types (regression coefficient=3.89, 95% CI −1.48 to 9.25, p=0.15). Self-reported pain intensity according to the FPS-R was most frequent in the moderate range (4–6; 51%), followed by low (29%) and high (19%).

Quality of life

There was no difference in self-reported quality of life for children and adolescents (5–18y; n=25) with or without chronic pain (z-score=0.75, p=0.45). However, carer proxy-reported (n=50) quality of life in children and adolescents (5–18y) was significantly reduced in those with chronic pain compared to those without (MD=1.03, 95% CI 0.80–13.27, p=0.03). For children (5–12y, n=45), there was no significant difference between self- or carer proxy-reported total quality of life scores (z-score=−1.47, p=0.14). However, carer proxy reporting for the pain and impact of disability domain was significantly lower compared to self-reporting

participants (z-score=−2.10, p=0.04). For adolescents (13–18y, n=30), there was no significant difference between self- or carer proxy-reported total quality of life scores (z-score=−1.68, p=0.09). However, carer proxy reporting was significantly lower for the communication and physical health (z-score=−2.39 p=0.02) and general well-being and participation domains (z-score=−2.86, p≤0.01) compared to self-reporting participants. No other significant differences were detected between the remaining self- and carer-reported domains for both children and adolescents.

DISCUSSION

This cohort of children and adolescents with predominant dyskinetic and mixed (dyskinetic/spastic) motor types, were heavily impacted by pain (85%), which was often chronic (77%). Almost half experienced pain that impacted their activity levels and was of moderate-to-high levels of intensity. Some body locations of pain were similar to sites previously identified across studies of broader CP cohorts (across all subtypes; e.g. lower limbs, abdomen), while others such as face, jaw, and temple pain were distinct to predominant dyskinetic motor types. Children and adolescents often experienced pain over multiple body locations, with increasing numbers of sites in those with more severe motor impairments. Chronic pain negatively influenced carer proxy-reported quality of life; however, it did not affect the quality of life of those able to self-report.

This study detected much higher pain and chronic pain prevalence in children and adolescents with predominant dyskinetic and mixed (dyskinetic/spastic) motor types compared to other published studies, inferring the possible risk for pain in this cohort. However, results need to be interpreted with caution considering the study was a non-population sample and the primary prevalence measure (HUI-3, 2wks) was carer-reported, which may contribute to overinflated findings. Compared to the findings of this study, much lower pain prevalence (46%) was reported in a Swedish population cohort (n=99) of predominant dyskinetic motor

types.[14] The lower pain prevalence in the Swedish study may be attributed to factors such as the use of no pain recall period and inclusion of younger children (<5y). However, a recent Australian prevalence study (n=280) undertaken by Ostojic et al. using similar measurements and sampling of a broad CP cohort, identified that predominant dyskinetic motor types were 3.5 times more likely and mixed (dyskinetic/spastic) motor types were 1.9 times more likely to have chronic pain compared to isolated spastic subtypes.[12] The risk for chronic pain highlighted by Ostojic et al. within the broader CP population, along with the high chronic pain prevalence detected in this study, highlight a need for lifespan pain management for populations with dyskinetic CP.

This study detected unique locations of bodily pain in children and adolescents with predominant dyskinetic and mixed (dyskinetic/spastic) CP motor types. The study findings suggest a possible trend for frequent upper-limb pain across the total study cohort (32%) compared to ranges of 4% to 19% identified across broader CP cohorts (all subtypes).[9] Frequent upper-limb pain may relate to the high levels of functional impairment detected in the cohort, recognized to be a risk factor across the broader CP population.[5] However, frequent upper-limb pain may also relate to bodily patterns of dystonia or choreoathetosis, with equal or more upper-limb body involvement detected in predominant dyskinetic compared to isolated spastic motor types.[38] Patterns of facial dystonia or choreoathetosis may have contributed to frequent face, jaw, and temple pain amongst predominant dyskinetic motor types.[10] Pain in this region may be caused by dystonia and/or choreoathetosis and a range of possible dental issues, such as dental disease, bruxism (habitual teeth grinding), traumatic dental injuries (e.g. biting), temporomandibular joint disorders, and mouth ulcers.[39,40] Interestingly, pain in the face, jaw, and temple has not been reported in other CP studies, which more broadly classify pain in this region as ‘head’ or ‘headaches’, which are commonly managed with simple analgesic medication.[38] Pain related to dystonia in the face, jaw, and temple may be specifically targeted

using treatments like botulinum neurotoxin A injections and gabapentin, along with dental treatments. Since individuals with a predominant dyskinetic motor type often have better eating and drinking ability compared to their high levels of motor impairment, pain in this region may have a more substantive impact on function and quality of life.[3] Ultimately, this study's findings highlight a need to pre-emptively target the diagnosis and treatment of pain in the face, jaw, and temple, and upper limbs, when managing pain in children with dyskinetic and mixed (dyskinetic/spastic) CP motor types, along with other common CP comorbidities.[41]

High numbers of body pain locations (on average five; GMFCS levels IV and V) were detected across the cohort, which increased at higher GMFCS levels. The presence of multiple body locations of pain may relate to total body involvement of CP, widespread bodily patterns of dystonia and/or choreoathetosis, and high frequency of medical comorbidities experienced by this population.[5] High numbers of body pain locations may also reflect the bias imposed by carers, who have the potential to overestimate the number of pain locations.[27] However, an alternative view is that the number of pain locations was actually underestimated in this study, given that the Childhood Arthritis and Rheumatology Research Alliance Body Diagram scores bilateral joints (e.g. the left and right knee) and some anterior/posterior body views (e.g. shin and calf) as one defined region.[31] In fact, the actual number of body regions where the presence of pain needed to be established was 59 rather than 21, making the Body Diagram quite laborious and fatiguing when administered to physically impaired augmentative and alternative communication users.[31] This led to some inconsistent performance and need to defer to carers, highlighting challenges in using generic pain tools in children with CP with severe physical and speech limitations who have the capacity to self-report. Furthermore, tools such as the FPS-R could not be completed in visually impaired individuals. Such challenges highlight the need to look towards adapting and psychometrically testing self-report generic tools to better

accommodate the needs of populations with dyskinetic CP, who are particularly vulnerable to not having their voices heard and reliance on carer proxy reporting in clinical practice.

Approximately half of the cohort with dyskinetic CP experienced moderate-to-high levels of pain intensity according to the PPP. Reasonably high pain intensity across the entire cohort with dyskinetic CP may align with trends for pain intensity to increase with the severity of motor impairment identified within broader CP cohorts.[9,38] Higher numbers of predominant dyskinetic motor types were identified to have moderate-to-high levels of pain intensity (PPP>14) compared to mixed motor types, but no differences were detected across the total PPP severity score. Use of the carer-reported PPP may impose some bias towards higher scores in predominant dyskinetic motor types, bearing in mind that 'involuntary movements', which are not necessarily painful, are embedded into the scoring system.[42]

This study achieved a moderate sample size (n=75) and response rate (59%) given the small proportional representation of children with dystonia or choreoathetosis within the CP population. However, the generalizability of study findings is limited by possible sampling and measurement bias. Selection bias may have positively skewed findings due to participant recruitment from specialist tertiary centres, implying the underlying complexity within the cohort. However, children with dyskinetic CP are characteristically complex, making tertiary recruitment appropriate in this circumstance. Positive skewing of results may also occur due to response bias, with those experiencing pain more likely to take part. There may also be some measurement error in recalling pain over 2 weeks for younger children. Ultimately, such biases highlight the complexity of undertaking pain research in dyskinetic CP populations, with the study results offering unique insight into these complexities and the pain features of children and adolescents with CP and dystonia or choreoathetosis. Future research needs to explore pain diagnosis and treatment in the face, jaw, and temple regions and evaluate methods for pain measurement for the unique population of augmentative and alternative communication users.

Conclusions

Children and adolescents with predominant dyskinetic and mixed (dyskinetic/spastic) CP motor types commonly experience pain that is often chronic and presents over multiple body locations, with some lesser recognized locations.

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Table 1: Baseline demographics of study cohort

Baseline measures	Total (n=75)	Mixed (n=38)	Dyskinetic (n=37)
Sex			
Male	44 (59)	21 (55)	23 (62)
Female	31 (41)	17 (45)	14 (38)
Age, y:mo, mean (SD)	10:11 (4:2)	10:7 (4:2)	11:4 (4:2)
Age group			
≤12y	45 (60)	24 (63)	21 (57)
>12y	30 (40)	14 (37)	16 (43)
Typology of CP			
Bilateral	69 (92)	36 (95)	33 (89)
Unilateral	6 (8)	2 (5)	4 (11)
GMFCS level			
I	6 (8)	3 (8)	3 (8)
II	6 (8)	4 (11)	2 (3)
III	7 (9)	5 (13)	2 (5)
IV	19 (25)	9 (24)	10 (27)
V	37 (49)	17 (45)	20 (54)
MACS level			
I	3 (4)	1 (3)	2 (5)
II	9 (12)	6 (16)	3 (8)
III	16 (21)	11 (29)	5 (14)
IV	13 (17)	6 (16)	7 (19)
V	34 (45)	14 (37)	20 (54)
CFCS level			
I	19 (25)	12 (32)	7 (19)
II	12 (16)	7 (18)	5 (14)
III	23 (31)	9 (24)	14 (38)
IV	9 (12)	4 (11)	5 (14)
V	12 (16)	6 (16)	6 (16)
Invasive tone management (ITB/DBS)	10 (13)	3 (8)	7 (19)
Previous bony hip surgery	29 (39)	16 (42)	13 (35)
Spinal fusion	11 (15)	8 (21)	3 (3)
Seizures	34 (45)	18 (47)	16 (43)
Gastrostomy	27 (36)	8 (21)	19 (52)
SEIFA[a]			
Quintile 1	10 (13)	6 (16)	4 (11)
Quintile 2	12 (16)	8 (22)	4 (11)
Quintile 3	10 (13)	5 (14)	5 (16)
Quintile 4	18 (24)	8 (22)	10 (26)
Quintile 5	25 (33)	11 (30)	14 (37)
Surgery 12 months prior	18 (24)	10 (26)	8 (22)

Data are n (%) unless otherwise stated. [a]Socio-Economic Indexes for Areas (SEIFA) Index of Relative Socio-Economic Disadvantage: quintile 1 describes the highest level of socio-economic disadvantage, whereas quintile 5 describes the lowest level. CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; CFCS, Communication Function Classification System; ITB, intrathecal baclofen; DBS, deep brain stimulation.

Table 2: Prevalence of Hypertonia Assessment Tool (HAT) scores (n=75)

HAT classification	Body region	Total (%)
Mixed	Left upper limb	48
	Right upper limb	52
	Left lower limb	77
	Right lower limb	81
Dystonic	Left upper limb	39
	Right upper limb	35
	Left lower limb	13
	Right lower limb	12

Table 3: Summary outcome measures, reporting, and missing data

Outcome measure	Mixed (n=38)	Dyskinetic (n= 37)
Chronic pain, n=75		
Carer-reported, 5–18y, n=57	29	28
Self-reported, 9–18y, n=14	8	6
Combination, 10–65y, n=4	1	3
Faces Pain Scale-Revised, n=31		
Self-reported, 5–18y	14	17
Body Diagram, n=75		
Carer-reported, 5–18y, n=41	21	20
Self-reported, 5–18y, n=29	15	14
Combination, 7–16y, n=5	2	3
Health Utilities Index-3, n=75		
Carer-reported, 5–18y	38	37
Paediatric Pain Profile, n=73		
Carer-reported, 5–18, n=73	36	37
Missing data, n=2	2[a]	0

[a]The PPP data for the first two participants were not collected.

Figure legends

Figure 1: Body locations of pain over the preceding 2 weeks in predominant dyskinetic and mixed (spastic/dyskinetic) motor types (n=75).

Figure 2: Number of body pain locations (mean and SD) according to Gross Motor Function Classification System (GMFCS) levels I–III (n=19) and IV and V (n=56).