





ORIGINAL ARTICLE

Pain trajectories and well-being in children and young people with cerebral palsy: A cohort study

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Abstract

Aim: To identify 5-week pain intensity trajectories and their association with physical and psychological well-being in children/young people with cerebral palsy (CP).

Method: A cohort study was conducted with 101 Canadian children/young people with CP, of whom 49 were female, with an overall mean age of 12 years 11 months (SD 3 years 1 month), range of 8 to 18 years, and classified in any Gross Motor Function Classification System level. Self-reported pain intensity (Faces Pain Scale – Revised) was collected weekly for 5 weeks and physical and psychological well-being (KIDSCREEN-27) at baseline and 5 weeks. Statistical analyses included latent class growth and general linear models.

Results: All Gross Motor Function Classification System levels were represented (I = 40.6%; II = 15.8%; III = 20.8%; IV = 13.9%; V = 8.9%). Five pain intensity trajectories were identified. Three trajectories had very low (35.4%), low (32.4%), or high (4.9%) mean stable pain. Two trajectories had moderate changing pain (16.8%) and high pain decreasing to moderate levels (10.5%) respectively. Trajectory participants with stable high pain had the lowest physical well-being (adjusted $\beta = -10.01$; 95% confidence interval [CI] = -19.37 to -0.66). Those in the three trajectories with the highest mean baseline pain intensity (>3 out of 10) had the lowest psychological well-being (adjusted $\beta = -8.27$, 95% CI = -14.84 to -1.70 ; $\beta = -6.74$, 95% CI = -12.43 to -1.05 ; $\beta = -5.82$, 95% CI = -15.34 to 3.71).

Interpretation: Almost one-third of participants had moderate-to-high pain intensity trajectories. Membership in the higher pain intensity trajectories was associated with lower physical and psychological well-being.

Cerebral palsy (CP) is a permanent, non-progressive disorder affecting movement and posture due to disturbances in the developing fetal or infant brain.¹ Despite the growing recognition of CP-related pain, little is known about the trajectories of pain in children/young people with CP and

their impact on well-being. Increased understanding could enable pain patterns to be identified, treatments targeted, pain chronicity minimized, and well-being enhanced.

Pain has been defined as ‘an unpleasant sensory and emotional experience associated with, or resembling that

Abbreviations: CTC, Children's treatment centre; FPS-R, Faces Pain Scale – Revised.

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associated with, actual or potential tissue damage.² Pain is one of the most commonly reported comorbidities in both children and adults with CP.³⁻⁶ In a recent systematic review of children and young adults, the reported prevalence of pain varied greatly, depending on the study, from 14% (any pain in the preceding month) to 76% (current pain using an observational/behavioural scale).⁷ Research has also identified that 25% of children with CP have moderate-to-severe chronic pain restricting daily activities.⁸

Evidence suggests there may be pain patterns experienced throughout childhood. In children with CP aged 3 to 16 years reporting pain at two visits, 26% had no pain, 20% had stable pain, and 21% had improved pain levels, while pain worsened in 33%.⁸ In a longitudinal study, Colver et al.⁹ also found that pain intensity increased from childhood to adolescence. Some individuals also experience significant long-term decreases in pain after procedures such as insertion of intrathecal baclofen pumps.¹⁰ Pain management in CP is challenging given the clinical heterogeneity of this population. The underlying aetiology, as well as access to and success of pain treatments, may ultimately influence short- and long-term pain trajectories.

To our knowledge, no previous studies have prospectively identified pain trajectories and their impact on the well-being and quality of life of children/young people with CP. Past work identified an association between pain and quality of life.^{9,11} Pain was associated with lower physical, psychological, and social well-being in children and adolescents.⁹ However, this study included only one follow-up period and pain trajectories were not identified. Another study found that pain was associated with health-related quality of life in children aged 3 to 19 years, with musculoskeletal deformity being the most important negative factor.¹²

The characteristics of individuals with CP, including age, sex, Gross Motor Function Classification System (GMFCS) level, CP subtype, sleep characteristics, and comorbidities such as mental health problems, are associated with pain and well-being.^{5,13,14} Furthermore, young females with CP are twice as likely to report more frequent and severe pain than their male counterparts.¹⁵ Other causes of pain that may diminish well-being include hip subluxation⁵ and CP management procedures, including wearing splints and botulinum neurotoxin A (BoNT-A) injections.¹³

Guided by gaps in the literature, stakeholder consultations, and our recent pilot study,¹⁶ we designed a cohort study. First, we aimed to identify short-term pain trajectories using self-reported pain intensity scores in a cohort of children and young people attending two Canadian children's treatment centres (CTCs). We hypothesized that pain follows one of five trajectories: increasing; decreasing; fluctuating; stable; and pain-free. Second, we aimed to determine whether pain trajectories were associated with short-term physical and psychological well-being. We hypothesized that compared to children/young people with no pain or decreasing pain trajectories, those reporting worsening, constant, or fluctuating pain trajectories would report lower physical and psychological well-being at 5 weeks.

What this paper adds

- Five distinct 5-week pain intensity trajectories were identified in children/young people with cerebral palsy.
- Thirty-two per cent of participants had moderate-to-high pain intensity trajectories.
- Participants in the trajectories with higher pain intensity reported lower physical and psychological well-being.

METHOD

Study design and settings

The University of Toronto and Holland Bloorview Kids Rehabilitation Hospital ethics boards approved the study. We conducted a cohort study in Ontario, Canada where all children diagnosed with CP are referred for assessment and/or care at their regional CTCs. We recruited from two CTCs: (1) Holland Bloorview Kids Rehabilitation Hospital (Holland Bloorview); and (2) Grandview Children's Centre (Grandview).

Study sample

We recruited participants between November 2019 and August 2020 who had CP, were 8 to 18 years old, were classified in any GMFCS level, were willing to report their pain, and successfully completed a sorting task (used to confirm their understanding of ratings on a scale). Those unable to communicate independently or complete an electronic questionnaire, with or without assistive devices or aid, were excluded.

Recruitment

The recruitment strategies described previously¹⁶ included: (1) posters/electronic signage at CTCs; (2) social media; and (3) study introduction to potential participants during outpatient clinic appointments by study staff or CTC physiotherapists. We also used a voluntary research call list at site 1 and mailed a study poster and telephoned potential participants using a list of CTC clients at site 2.

We used a two-step screening process to determine eligibility. First, we contacted clients/parents by telephone to introduce the study, complete an initial telephone screen (identifying clients 8–18 years old, diagnosed with CP, and able to communicate adequately to complete the questionnaires), and book an in-person meeting. In March 2020, in-person data collection was halted in compliance with COVID-19 pandemic precautions. We conducted the

screening and baseline meetings using a secure, hospital-based virtual platform from April 2020 onwards. If eligible and interested, parents/guardians and children/young people were asked to provide written, informed consent and/or assent to participate.

Data collection

Standardized self-report baseline and follow-up questionnaires were circulated and completed using the REDCap data collection tools.¹⁷ Follow-ups were completed using electronic questionnaires at 7, 14, 21, 28, and 35 days post-baseline. Figure S1 presents the data collection processes, including pre-study stakeholder engagement and a pilot study.¹⁶ Additionally, data on self-reported CP-related care in the past month was collected at baseline and at the 5-week follow-up.

Exposure: pain trajectory group membership

We used the Faces Pain Scale – Revised (FPS-R) once weekly for 5 weeks as the primary scale for measuring pain intensity in the past week to identify short-term pain intensity trajectories.¹⁸ This questionnaire is recommended for children/young people aged between 7 and 18 years and is suitable for those with limited numeracy skills.¹⁹ It includes six sex-neutral faces depicting ‘no pain’ to ‘most pain possible’ ordered numerically from 0 to 10.²⁰ It has adequate reliability, validity (construct and content), and responsiveness.¹⁹

Outcome: physical and psychological well-being

Using the KIDSCREEN-27 physical and psychological well-being domains, we assessed well-being in the preceding week at baseline and at the 5-week follow-up. KIDSCREEN-27 items are scored on a 5-point Likert scale (1 = not at all to 5 = very much); it is applicable to typically developing and chronically ill children aged 8 to 18 years, has acceptable internal consistency and test–retest reliability, and has been used in various settings and with children with CP.^{9,21,22} The domains were reported as group T scores, allowing comparison with an international reference population (mean = 50, SD = 10).²¹ Some participants had the option to complete a shorter version of the questionnaire, excluding the KIDSCREEN-27, if it was deemed too burdensome based on individual ability.

Potential confounders

Potential confounders were identified based on the previous scientific literature and clinical experience. These included sex, age, GMFCS level, CP subtype, sleep disturbances, mental health conditions, and socioeconomic status.

Sociodemographic data

At baseline, age, sex, and socioeconomic status (postal code linked with median household income census data) were recorded.^{23–25}

Clinical data

CP subtype, GMFCS level, mental health disorders, and other comorbid conditions (i.e. epilepsy and seizure disorders, gastrointestinal conditions) were collected from electronic health records.^{26,27} Sleep characteristics were collected using the PROMIS Sleep Disruption Questionnaire (Short Form, v1.0-4a) and were reported as standardized T scores.²⁸

Sample size

We estimated sample size using the method of effect size for latent class analysis and considered scenarios based on the different numbers of trajectories and distribution of participants across trajectories.²⁹ The analysis was powered for the likelihood ratio test comparing a $k-1$ model solution to a k model solution. For power = 0.80 and $\alpha = 0.05$, a sample size of 90 to 100 participants was estimated to be adequate to identify five pain trajectories based on the tables by Dziak et al.²⁹ produced using Monte Carlo simulations.

Statistical analysis

Descriptive statistics were reported using means, medians, and proportions.

Objective 1

Latent class growth modelling³⁰ was used to identify distinct pain trajectories. Participants were assigned to a single trajectory based on the highest probability of trajectory membership. Latent class models were built successively, starting with a one-cluster model. Each model began by including cubic, quadratic, and linear terms. Each successive model added a trajectory group, up to $k = 5$. The optimal model was decided by comparing and choosing models with the lowest criterion value: (1) the Bayesian information criterion; and (2) the Akaike information criterion. To ensure parsimony, we reduced the parameters in the model by removing cubic, quadratic, and linear terms that were not significantly different from zero. Spaghetti plots for participants assigned to each trajectory from the final model were created to examine individual variation within the trajectories (Figures S2–S6).

Objective 2

We used general linear models to measure the crude association between short-term pain trajectories and well-being at 5 weeks. Bivariate models were constructed with potential confounders. Any confounders meeting the change in estimate approach ($\pm 10\%$ change from the crude model) were included in the final multivariable models.³¹

Sensitivity analyses

We performed sensitivity analyses with latent class growth modelling using the first 4 weeks of pain measurement to assess if the cross-sectionality of the measurement of pain and well-being done on the same day influenced trajectory shape and group membership. We also assessed if models were sensitive to individuals with no pain by removing pain-free participants.

We used the SAS software v9.4 (SAS Institute, Cary, NC, USA). We downloaded the traj application³² from <http://www.andrew.cmu.edu/user/bjones/>.

RESULTS

We identified 550 eligible participants and needed to contact 260 to recruit 100 (Figure S7). At the stage 1 screening, of the 260 individuals who were contacted, 190 were eligible to attend in-person screening. At the stage 2 screening, 111 were screened in-person. Overall, 53.7% (102 out of 190) met the inclusion criteria and consented to participate. One child withdrew before completing the baseline questionnaire. The follow-up rate was above 90% at all time points. Eight participants did not complete the KIDSCREEN-27 at 5 weeks (three withdrew after baseline and five completed the short-form questionnaires) resulting in 93 responses for the final KIDSCREEN-27. There were few missing data; therefore, imputation was not performed. The ages of participants ranged from 8 to 18 years and 48.5% were female (Table 1). Mean baseline pain intensity (FPS-R) was 2.8 (2.6) and 76.2% reported experiencing pain in the preceding week. Eighty percent reported doing CP-related home exercises in the preceding month, 67.3% wore a brace, 24.8% had physiotherapy, 17.8% did yoga, and 9% had massage therapy. Pharmacological treatments potentially resulting in some pain management (BoNT-A, gabapentin, omeprazole, cannabidiol oil) varied by trajectory group and included: group 1 = 10 out of 34 (BoNT-A only); group 2 = 11 out of 34 (BoNT-A only); group 3 = 10 reports among 17 individuals (BoNT-A = 7, gabapentin = 2, cannabidiol oil = 1, not exclusive to each person); group 4 = 7 reports among 11 individuals (BoNT-A = 4, gabapentin = 1, omeprazole = 2, not exclusive to each person); and group 5 = 2 out of 5 (BoNT-A only). A variety of reported mental health disorders or symptoms and developmental/behavioural disorders were reported in the electronic health records. These included 17 reports of

anxiety, seven of depression, 15 of attention-deficit/hyperactivity disorder/attention-deficit disorder, five of autism spectrum disorder, two of obsessive-compulsive disorder, and one each of bipolar disorder, oppositional defiance disorder, high stress, and self-harm behaviours. Potential sources of pain included constipation ($n = 19$), gastro-oesophageal reflux ($n = 7$), presence of a gastrostomy tube ($n = 3$), epilepsy/seizure disorders ($n = 11$), and other comorbid conditions including scoliosis ($n = 9$), reactive airway disorder ($n = 2$), ventriculoperitoneal shunt ($n = 2$), and kidney stones ($n = 1$).

Pain intensity trajectories

The optimal number of trajectories resulting from latent class growth modelling was five (Figure 1). Trajectories 1, 2, and 5 used intercept terms only (constant average pain over time), while trajectories 3 and 4 contained both quadratic and linear terms. The highest probability of participant assignment to an individual trajectory ranged between 0.85 and 1.0, indicating distinct classification. This model had the lowest Bayesian information criterion (-1227.05) and Akaike information criterion (-1208.74) (Table S1). The final model trajectories are described as: trajectory 1: no or very mild stable mean pain ($n = 34$, 35.4%), with a constant pattern and estimated mean weekly pain of 0.69 (95% confidence interval [CI] = 0.24–1.15); trajectory 2: mild stable mean pain ($n = 34$, 32.4%), with a constant pattern and estimated mean pain intensity of 1.99 (95% CI = 1.49–2.49); trajectory 3: moderate changing mean pain ($n = 17$, 16.8%), with moderate pain intensity at baseline, plateauing to above 5 out of 10 at weeks 2 and 3, then decreasing below the mean estimated baseline level at week 5 (3.93, 95% CI = 3.12–4.75); trajectory 4: high to decreasing moderate mean pain ($n = 11$, 10.5%) with high mean pain intensity at baseline (estimated mean = 7.47 95% CI = 6.32–8.62) improving over time (week 5 estimated mean = 3.25, 95% CI = 2.16–4.33); trajectory 5: high stable mean pain ($n = 5$, 4.9%), with a constant pattern and estimated mean of 7.33 (95% CI = 6.54–8.11).

Individual participant variability within each trajectory group was visualized with spaghetti plots (Figures S2–S6) and mostly followed the shape of the assigned group.

Physical and psychological well-being

Trajectory group membership was associated with both physical and psychological well-being in the crude model (Table 2). Individuals in trajectory group 1 reported the highest physical well-being. In the crude model, there was a downward gradient of physical well-being based on trajectory group membership. Compared to group 1 with a mean physical well-being (model intercept) of 55.31 (95% CI = 52.03–58.78), members in all other trajectory groups had lower mean physical well-being. We used the change in estimate approach to construct

TABLE 1 Baseline characteristics of the study participants ($n = 101$)

Characteristic	Full sample ($n = 101$)	Trajectory group 1 ($n = 34$)	Trajectory group 2 ($n = 34$)	Trajectory group 3 ($n = 17$)	Trajectory group 4 ($n = 11$)	Trajectory group 5 ($n = 5$)
Age, years:months						
Mean (SD)	12:10 (3:1)	13:3 (3:3)	12:8 (3:1)	13:1 (2:10)	11:10 (3:7)	13:2 (2:7)
Median (IQR)	13:0 (6)	13:0 (5)	12:0 (6)	13:0 (5)	11:0 (7)	13:0 (2)
Female, n (%)	49 (48.5)	11 (32.4)	19 (55.9)	12 (70.6)	3 (27.3)	4 (80.0)
CP type, n (%)						
Spastic	69 (68.3)	27 (79.4)	22 (66.7)	12 (70.6)	4 (36.4)	4 (80.0)
Dyskinetic	13 (12.9)	1 (2.9)	4 (12.1)	3 (17.7)	5 (45.5)	0
Mixed (spastic and dyskinetic)	14 (13.9)	6 (17.7)	3 (9.1)	2 (11.8)	2 (18.2)	1 (20.0)
Ataxic	3 (3.0)	0	3 (9.1)	0	0	0
Unknown	2 (2.0)	0	1 (3.0)	0	0	0
GMFCS level, n (%)						
I	42 (41.6)	18 (52.9)	14 (41.2)	6 (35.3)	2 (18.2)	2 (40.0)
II	15 (14.9)	3 (8.8)	5 (14.7)	5 (29.1)	2 (18.2)	0
III	21 (20.8)	6 (17.7)	9 (26.5)	3 (17.7)	2 (18.2)	1 (20.0)
IV	14 (13.9)	7 (20.6)	4 (11.8)	0	1 (9.1)	2 (40.0)
V	9 (8.9)	0	2 (5.9)	3 (17.7)	4 (36.4)	0
Average pain intensity (FPS-R) ($n = 100$)						
Mean (SD)	2.8 (2.6)	0.9 (1.3)	2.0 (1.7)	3.7 (1.3)	7.5 (1.6)	6.8 (1.1)
Median (IQR)	2.0 (4)	0 (2)	2.0 (2)	4.0 (2)	8.0 (2)	6.0 (2)
KIDSCREEN-27, mean T score (SD)						
($n = 96$)		($n = 33$)	($n = 32$)	($n = 16$)	($n = 9$)	
Physical well-being	50.0 (11.1)	52.2 (10.3)	49.7 (7.4)	47.4 (12.4)	52.8 (19.9)	39.9 (5.9)
Psychological well-being	51.0 (10.1)	53.8 (10.5)	49.6 (7.7)	45.6 (8.7)	51.8 (10.4)	41.7 (5.9)

Abbreviations: CP, cerebral palsy; FPS-R, Faces Pain Scale – Revised; GMFCS, Gross Motor Function Classification System; IQR, interquartile range.

multivariable models and controlled for baseline physical well-being, GMFCS level, CP subtype, and median household income in the final physical well-being model (Table 2). The gradient remained but associations were weaker for all groups. Compared to trajectory group 1, all other groups had lower estimates; membership in trajectory 5 was associated with the largest difference in physical well-being from trajectory 1 ($\beta = -10.01$, 95% CI = -19.37 to -0.66).

In the psychological well-being crude model, those in groups 3 and 5 had the lowest mean psychological well-being compared to group 1 ($\beta = 56.40$, 95% CI = 53.07 – 59.74) (Table 2). In the adjusted model, we controlled for baseline psychological well-being, mental health, sleep disturbance, and median household income (Table 2). The difference in estimated mean psychological well-being compared to group 1 was almost unchanged for members in groups 2 and 4, while the association was weaker for those in groups 3 and 5 when confounders

were controlled. Compared with group 1, membership in trajectory groups 3 (-6.74 ; 95% CI = -12.43 to -1.05), 4 (-8.27 ; 95% CI = -14.84 to -1.70), and 5 (-5.82 ; 95% CI = -15.34 to 3.71) was associated with lower mean psychological well-being.

Sensitivity analyses

The cross-sectional measurement of pain and well-being did not bias the results. Pain reported at 5 weeks did not influence trajectory membership compared to trajectory membership at 4 weeks, with 74 out of 101 participants in the same trajectory groups in both FPS-R models. The trajectory shape varied slightly between the 4- and 5-week models. When excluding participants who reported no pain ($n = 13$), there were some changes in group membership but the overall trajectory shapes were the same (Appendix S1).

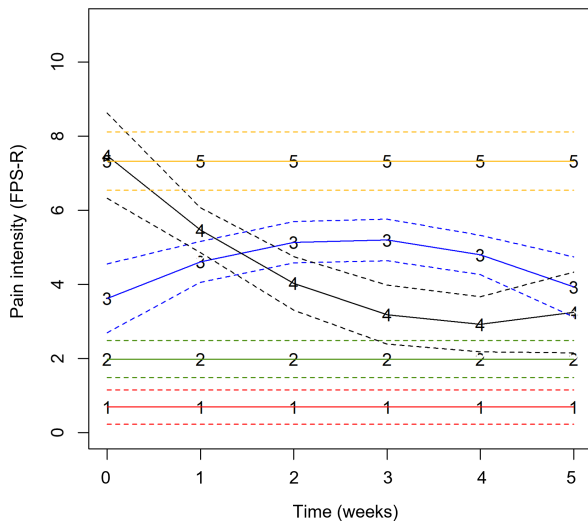


FIGURE 1 Model for trajectory group membership for pain intensity measured by the Faces Pain Scale - Revised (FPS-R) over 5 weeks ($n = 101$). The dashed lines represent the 95% confidence intervals. Group percentages: 1 = 35.4%, 2 = 32.4%, 3 = 16.8%, 4 = 10.5%, 5 = 4.9%

DISCUSSION

This study yielded new findings regarding the clinical course of pain intensity in children/young people with CP. This was conceptualized as the pain intensity for any type of pain experienced in the previous week, whether related to their CP or not. The short-term pain trajectory model describes five distinct groups. Three groups had stable trajectories with distinct mean pain intensities. Of these, two had participants with no or low stable mean pain intensity (groups 1 and 2), representing 67.8% of participants; the third (group 5) had participants (5%) with high stable mean pain intensity. Members of trajectory group 3 (16.8%) had changing moderate intensity pain. This suggests they may have had more fluctuating pain intensity, which would need to be confirmed with longer follow-ups. Those in trajectory group 4 (10.5%) had high initial pain that decreased over time. It is disconcerting that approximately one-third of children were in trajectories with moderate-to-high pain intensity levels.

To our knowledge, this is the first study to identify short-term pain intensity trajectories in children with CP. Past research assessed how pain changes in children with CP using two time points spanning several months to years.^{8,9} By characterizing short-term pain trajectories in children with CP, future studies may focus on identifying factors that have an impact on or improve pain trajectories. Studies of pain trajectories in other paediatric chronic conditions, such as rheumatoid arthritis and sickle cell anaemia, reported similar results.^{33,34} Children with juvenile rheumatoid arthritis also grouped into concerning pain trajectories, with chronically moderate and increasing or decreasing pain trajectories; however, these were measured every 6 months for 5 years.³³ Children hospitalized for sickle cell vaso-occlusive crises experienced slow, moderate, and rapidly decreasing pain trajectories over 1 week.³⁴

The pain intensity trajectory model presented in this study is not based on an inception cohort. Instead, it provides a 5-week snapshot of pain trajectories and serves as a starting point for future work. Participants in the high and moderate pain intensity trajectories had lower physical well-being compared to those in the low pain intensity trajectories; the effect was larger the higher the pain intensity. Participants in the trajectories with higher mean baseline pain intensities and changing pain intensities had lower psychological well-being compared to those in the stable, lower pain intensity trajectories. Patterns in the crude models, such as lower psychological well-being in trajectory groups 3 to 5 compared to group 1, were maintained although diminished in the adjusted models. Children in lower pain trajectories may experience less impact on daily activities leading to higher psychological well-being. Furthermore, some children may cope better with stable pain levels than unanticipated changes or fluctuations. Also, given that children/young people in three trajectory groups had constant pain levels, there may be a cross-sectional relationship with well-being versus an association with the trajectories. However, this was tested in our sensitivity analyses and any impact was minimal.

Strengths of the current study include frequent follow-ups in a short time period, control of confounders, low attrition, and validated questionnaire instruments. Questionnaires were self-completed and participants were classified in all GMFCS levels, CP subtypes, and a wide age range. Our study has some limitations. First, uncertainties remain regarding the psychometric properties of the FPS-R when used in children/young people with chronic pain; it is possible we may be assessing chronic versus acute pain episodes.¹⁹ Second, the small sample size of some trajectory groups led to greater variability around the point estimates. Future work to identify pain trajectories should ensure a sufficient sample size for each trajectory group, thereby leading to greater statistical power. Third, misclassification bias is possible by using the postal code as an indicator for income and socioeconomic status. Fourth, we attempted to be inclusive of all GMFCS levels by including participants who could not verbalize (but could vocalize, or use eye gaze or communication devices) or complete questionnaires independently (could direct someone else to fill in their electronic responses). However, we excluded some individuals based on their inability to complete a sorting task and individuals in GMFCS levels IV and V may be over-represented in this group. Thus, our results may not be representative of individuals with lower functional status or severe comorbidities. Fifth, we did not consistently record limb involvement during data collection, which is a limitation because this helps to better assess the generalizability of results to specific CP subtypes. Finally, it is probable that residual confounding exists in our well-being models.

Some of the trajectory shapes differed from our hypotheses. No trajectories had increasing pain or no pain. Although we measured pain over a short period, future work should assess if trajectory patterns change over longer periods (e.g. up to 1 year), measure ethnic, cultural, and sex differences,

TABLE 2 Association between trajectory group membership (FPS-R) and physical and psychological well-being (KIDSCREEN-27), controlling for confounders

Variable	Crude models (<i>n</i> = 93)		Adjusted models (<i>n</i> = 91)	
	β^a (SE)	95% CI	β^a (SE)	95% CI
Physical well-being				
Group 1 (ref)	55.31 (1.65)	52.03 to 58.78	33.79 (5.02)	23.81 to 43.77
Group 2	-6.19 (2.38)	-10.93 to -1.46	-4.48 (2.30)	-9.06 to 0.09
Group 3	-8.35 (2.97)	-14.26 to -2.44	-6.76 (2.71)	-12.16 to -1.36
Group 4	-8.42 (3.60)	-15.57 to -1.28	-7.87 (3.41)	-14.65 to -1.08
Group 5	-16.30 (5.07)	-26.38 to -6.22	-10.01 (4.70)	-19.37 to -0.66
Baseline physical well-being	-	-	0.41 (0.09)	0.24 to 0.58
GMFCS levels I and II (ref)	-	-	-	-
GMFCS levels III and IV	-	-	-2.04 (1.94)	-5.90 to 1.82
CP subtype - spastic (ref)	-	-	-	-
CP subtype - other ^b	-	-	-1.11 (2.06)	-5.21 to 3.00
Household income (ref) \geq Can\$100 000 ^c	-	-	-	-
Household income Can\$75 000 to <\$100 000 ^c	-	-	1.03 (2.39)	-3.71 to 5.77
Household income <Can\$75 000 ^c	-	-	2.68 (2.27)	-1.83 to 7.19
<i>R</i> ²	0.17		-	
Psychological well-being				
Group 1 (ref)	56.40 (1.68)	53.07 to 59.74	32.44 (5.38)	21.74 to 43.14
Group 2	-3.31 (2.43)	-8.13 to 1.52	-3.25 (2.30)	-7.82 to 1.33
Group 3	-10.53 (3.03)	-16.55 to -4.50	-6.74 (2.86)	-12.43 to -1.05
Group 4	-8.40 (3.67)	-15.69 to -1.11	-8.27 (3.30)	-14.84 to -1.70
Group 5	-12.38 (5.17)	-22.65 to -2.10	-5.82 (4.79)	-15.34 to 3.71
Baseline psychological well-being	-	-	0.43 (0.10)	0.24 to 0.61
Mental health disorder: none reported (ref)	-	-	-	-
Mental health disorder: reported	-	-	-0.71 (2.34)	-5.38 to 3.95
Sleep disturbance: lower median T score \leq 55.8 (ref) ^c	-	-	-	-
Sleep disturbance: higher ^d Median T score > 55.8	-	-	-2.41 (1.93)	-6.25 to 1.44
Household income (ref) \geq Can\$100 000 ^c	-	-	-	-
Household income Can\$75 000 to <Can\$100 000 ^c	-	-	2.97 (2.38)	-1.77 to 7.70
Household income <Can\$75 000 ^c	-	-	3.46 (2.25)	-1.02 to 7.93
<i>R</i> ²	0.16		-	

^a β , unstandardized parameter estimate. ^bIncludes dyskinetic, mixed, ataxic, and unknown. ^cMedian household income using the 2016 Canada census data. ^dPROMIS Sleep Disturbance Short Form Questionnaire 4a. Abbreviations: CI, confidence interval; CP, cerebral palsy; FPS-R, Faces Pain Scale - Revised; GMFCS, Gross Motor Function Classification System; ref, reference group; SE, standard error.

be repeated in other jurisdictions to assess generalizability, and importantly include qualitative data collection to help identify pain triggering and relieving factors. The next step of our work will be to analyse how pain interference, a more complex and multidimensional construct, is associated with both pain and well-being scores. Continued measurement of socioeconomic status of individuals with CP is important. This was a confounder in both the physical and psychological well-being models. Lower socioeconomic status is associated with higher level of motor impairment (GMFCS), intellectual impairment, and additional comorbidities.³⁵

Furthermore, those who are economically and socially disadvantaged may have greater difficulty accessing care for their children, which may have an impact on pain experiences and well-being. Health assessments could also help determine pain aetiology. Identifying not only pain intensity but its aetiology and types of pain management will be important in future work to better understand pain trajectories. For instance, many children and young people with hip joint displacement may experience significant and prolonged pain. Equally, children and young people with CP who have dystonia, muscle contractures, or experience

constipation may have very different pain trajectories than those who do not have these conditions. This is especially true for children and young people in GMFCS levels IV and V who often experience multiple sources of pain. Lastly, future work should consider the use of proxies to respond on behalf of children/young people with CP with lower functional ability or cognitive impairments. Proxies can provide important information regarding pain and well-being for which there is currently little reported in this subgroup.

Implications

Children with CP often have repeated exposure to acute pain resulting from daily activities and various procedures including BoNT-A injections, bracing, casting, and therapy.¹³ Without adequate knowledge of how pain changes in the short term, effective treatments may not be provided to ameliorate the long-term consequences of pain. It is important to identify and minimize acute pain to diminish the negative impact on physical and psychological well-being and prevent pain hypersensitization. Neuroplastic changes, either functional or structural, have the negative effect of reinforcing pain and contributing to pain chronicity.³⁶ If pain limits function and/or becomes chronic, individuals may be at risk for chronic pain in adulthood, having diminished well-being, restriction in participation, and greater incidence of mental health symptoms. Repeated pain assessments may identify those at risk for concerning pain trajectories and lead to (1) the identification of successful pain prevention interventions, (2) prevention of pain chronicity, and (3) maintaining or improving well-being.

CONCLUSION

This work identified five pain trajectories among children/young people with CP and established that for 68% the estimated mean pain was predominantly stable over 5 weeks. There is an association between trajectory groups with high stable pain intensity and lower physical well-being. Individuals in the trajectory groups with higher baseline pain intensity had lower psychological well-being. Future research should include longer-term follow-ups and assess the impact of pain trajectories on participation.

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

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DATA AVAILABILITY STATEMENT

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

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SUPPORTING INFORMATION

The following additional material may be found online.

Table S1: Goodness of fit indices for the tested trajectory models, $k = 1$ to $k = 5$, for cubic, quadrilateral, linear, and mixed terms.

Figure S1: Schematic of the study cohort.

Figure S2: Spaghetti plot demonstrating FPS-R score variability in trajectory group 1 for participants reporting pain at one or more time points ($n = 34$).

Figure S3: Spaghetti plot demonstrating FPS-R score variability in trajectory group 2 for participants reporting pain at one or more time points ($n = 34$).

Figure S4: Spaghetti plot demonstrating FPS-R score variability in trajectory group 3 for participants reporting pain at one or more time points ($n = 17$).

Figure S5: Spaghetti plot demonstrating FPS-R score variability in trajectory group 4 for participants reporting pain at one or more time points ($n = 11$).

Figure S6: Spaghetti plot demonstrating FPS-R score variability in trajectory group 5 for participants reporting pain at one or more time points ($n = 5$).

Figure S7: STROBE flow chart for study recruitment, enrollment, and follow-up.

Appendix S1: Sensitivity analyses.

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