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Original Article

Health-related quality of life in Canadian children with cerebral palsy: what role does sleep play?

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ABSTRACT

Objective: To evaluate, in Canadian children with cerebral palsy (CP): (1) health-related quality of life (HRQoL) as well as (2) associations between HRQoL and (a) sleep problems, (b) nighttime pain, and (c) child characteristics (eg, age, CP phenotype, comorbidities).

Methods: Children aged 3–12 years were recruited from neurology clinics and a provincial CP registry. Caregivers completed the Pediatric Quality of Life Inventory (PedsQL) Generic Core and CP Modules as well as the Sleep Disturbance Scale for Children (SDSC) to assess HRQoL and sleep, respectively. Child characteristics were extracted from hospital records and registry data.

Results: A total of 146 children with CP (mean age ± standard deviation: 6.9 ± 2.9 years) completed the study. Impaired HRQoL (scores more than 2 SDs below the normative population mean) on the PedsQL Total, Physical and Psychosocial Health scales was found in 33.6%, 38.4% and 17.6% of children, respectively. Non-ambulatory status, sleep problems and significant comorbidity were the strongest predictors of impaired Total HRQoL, with odds ratios (95% confidence intervals) of 30.1 (8.2–110.4), 3.8 (1.1–12.5) and 3.3 (1.2–9.2), respectively, adjusted for young age (5–7 years) and nighttime pain. Non-ambulatory status and sleep problems exclusively increased the risk of impaired physical and psychological health, respectively, with adjusted ORs (95% CIs) of 58.3 (11.9–284.5) and 7.5 (2.5–22.5). More severe sleep problems were associated with worse pain-related HRQoL.

Conclusions: Sleep, non-ambulatory status and presence of comorbidities are important determinants of HRQoL in children with CP. Monitoring sleep, psychosocial functioning and pain is important in this population, as their management should improve HRQoL.

1. Introduction

Sleep disturbances are common in children with neurodevelopmental disorders [1]. Children with cerebral palsy (CP), the most common cause of pediatric physical disability [2], are at risk for significant sleep problems. In addition to the severity of the primary motor impairment [3–6], comorbidities, such as severe visual [7], auditory [7], cognitive impairment [3,4], and/or epilepsy [3,4,8], as well as significant pain [9] from spastic limbs, musculoskeletal deformity and/or interventions have been reported to be important determinants of sleep disturbances in children with CP.

Health-related quality of life (HRQoL) is a clinically important outcome for research and interventions targeting children with neurodevelopmental disorders, including CP. HRQoL involves the physical, mental and social aspects of an individual's health, and an assessment of their well-being and/or functioning within these domains of the construct [10]. Varni and colleagues [11] were among the first to measure HRQoL in children with CP, reporting significantly lower physical, emotional, social and school functioning scores in children with CP compared with their typically-developing peers. The same researchers [12] later reported that

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Abbreviations: CP, cerebral palsy; DIMS, disorders of initiation and maintenance of sleep; HRQoL, health-related quality of life; PedsQL, Pediatric Quality of Life Inventory; SDSC, Sleep Disturbance Scale for Children.

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among 33 pediatric disease conditions, children with CP had the lowest HRQoL, a finding that underscores the importance of assessing child characteristics that could negatively affect HRQoL and be potential targets for interventions in children with CP.

Of note, many of the same risk factors known to put children with CP at increased risk for sleep problems have been reported to be important determinants of HRQoL; these factors include a greater degree of motor impairment [13–16], presence of comorbidities [16], older age [17] and pain [14,16,17]. To date, the possible role of sleep problems as a determinant of HRQoL has been largely understudied. Children with CP and sleep disordered breathing who underwent treatment with adenotonsillectomy were shown to have had significant improvements in HRQoL postoperatively [18]. Sandella and colleagues [19] found that caregiver-reported symptoms of daytime sleepiness and insomnia negatively impacted physical and psychosocial dimensions of HRQoL, respectively, in a small sample (n = 41) of school-aged children with CP. Most recently, Zuculo and colleagues [20] found, in a small sample (n = 43) of four to 18-year-olds with CP, several significant negative correlations between certain aspects of HRQoL and sleep disorders. Most notably, excessive daytime sleepiness was negatively associated with decreased physical function, as well as with overall health, behavior, well-being and self-esteem. Disorders of initiation and maintenance of sleep (which can include symptoms of insomnia) were associated with decreased physical function and self-esteem. Given the previously limited sample sizes, it remains unclear if sleep problems contribute to impaired HRQoL in children with CP within specific age groups (preschool-vs school-aged) and to what degree, if any, sleep problems may impact HRQoL when also accounting for the effects of other known risk factors for impaired HRQoL.

Our study aimed to evaluate, in a large sample of Canadian preschool- and school-aged children with CP, the relationship between HRQoL and sleep problems, pain and child characteristics, including age, CP phenotype, and comorbidities. Our hypotheses were that: (1) a large percentage of children would have impaired HRQoL as assessed on the Pediatric Quality of Life Inventory (PedsQL) Generic Core Module; (2) impaired HRQoL would be associated with a more severe CP phenotype, greater comorbidity, and the presence of sleep problems, as assessed on the Sleep Disturbance Scale for Children (SDSC); and (3) HRQoL related to pain (as assessed on the PedsQL Cerebral Palsy Module) would be associated with a greater severity of sleep problems on the SDSC.

2. Methods

2.1. Participants

As part of a broader cross-sectional study investigating the prevalence of sleep problems in children with CP [21], 150 children were prospectively recruited through hospital neurology clinics and a provincial CP Registry. Inclusion criteria for this larger study were: age 3–12 years; diagnosis of CP by a neurologist; and living at home with a primary caregiver or guardian who is fluent in French or English. For the current study, we excluded children with incomplete HRQoL data (n = 4). The study protocol was approved by the Research Ethics Board of the Montreal Children's Hospital and participating caregivers provided written informed consent to the research and publication of results.

2.2. Data collection

Caregivers completed two questionnaires about HRQoL and two questionnaires about sleep, as detailed below.

2.2.1. Health-related quality of life measures

Caregivers of children with CP completed two HRQoL measures: 1) the Pediatric Quality of Life Inventory (PedsQL) 4.0 Generic Core Parent-proxy Module [22], designed for use in healthy children as well as children with chronic health conditions; and 2) the PedsQL 3.0 Cerebral Palsy Parent-proxy Module [23], designed to measure HRQoL dimensions specific to children with CP.

2.2.1.1. PedsQL Generic Core Module. The PedsQL Core Module is comprised of 23 items divided into the following subscales: (1) Physical Functioning (eight items), (2) Emotional Functioning (five items), (3) Social Functioning (five items), and (4) School Functioning (five items). Caregivers completed the version of the questionnaire that corresponded to the age of their child [3–4 years ('toddler'), 5–7 years ('young child'), or 8–12 years ('child')]; items for each version differed only in terms of age-specific language, but not in content. The scale assesses caregivers' perceptions of their child's HRQoL by asking how much of a problem each item has been for the child during the last month; responses are provided on a 5-point scale (0 = 'never a problem', 1 = 'almost never a problem', 2 = 'sometimes a problem', 3 = 'often a problem', 4 = 'almost always a problem'). Items were reverse scored and linearly transformed to a 0–100 scale (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0), with higher scores indicating better HRQoL. Scale scores were computed as the sum of completed items divided by the number of items answered. If >50% of the items were missing, the scale/subscale score was not computed (explaining the differences in 'n' for the scales/subscales). The PedsQL Generic Core Module yields a Total score (sum of all items) which is subdivided into a Physical Health summary score (same as the Physical Functioning subscale) and a Psychosocial Health summary score, which is the sum of the remaining 15 items on the Emotional, Social, and School Functioning subscales divided by the number of items answered. A large population-based study of more than 10,000 children [24] has provided normative means and standard deviations (SDs) for the scale and subscale scores; children with scores more than 1 SD below the population mean were said to represent an at-risk status for impaired HRQoL in the study. In the current study, we determined both the number of children who were 1 SD and 2 SDs below the population mean. As we were interested in examining the factors associated with children with CP with more impaired HRQoL, we defined children with scores more than 2 SDs below the population mean as having impaired HRQoL.

2.2.1.2. PedsQL Cerebral Palsy Module. The PedsQL CP Module is comprised of 35 items divided into the following seven subscales: (1) Daily Activities (nine items), (2) School Activities (four items), (3) Movement and Balance (five items), (4) Pain and Hurt (four items), (5) Fatigue (four items), (6) Eating Activities (five items) and (7) Speech and Communication (four items). The version for toddlers is comprised of 22 items; there are no School Activities and Speech and Communication subscales, and the Daily Activities and Eating Activities subscales contain fewer items. The PedsQL CP Module uses the same 5-point scale and has the same scoring procedures as the Generic Core Module. The PedsQL CP Module is different in that it provides only subscale scores (no total or summary scores), and the number of items differs with the age version used. As there are currently no normative population means for this measure, scores on the PedsQL CP Module subscales were used as continuous variables in our study.

2.2.2. Sleep problems

The Sleep Disturbance Scale for Children (SDSC) is a screening tool used to assess the most common symptoms of pediatric sleep disorders. The SDSC was initially validated for use in children and youth 6–18 years [6] and more recently, for use in preschool children aged 3–5 years [25]. Caregivers are asked to report the frequency of 26 sleep items on a 5-point scale from ‘never’ to ‘daily’. The SDSC yields a total score and score for six sleep disorders; we have previously published data on the prevalence of sleep disorders in this sample [21]. Raw scores can be converted to T-scores based on published norms for children [6,25] and can then be classified as normal (T-score $\leq$ 70) or abnormal (T-scores > 70). Abnormal total scores, which indicate clinically concerning sleep problems, were included as a predictor variable for impaired HRQoL in our univariate and multivariate analyses. To further explore the relationship between the 26 specific sleep items reported on the SDSC and HRQoL, we dichotomized responses on each SDSC item to classify if the sleep issue was a problem for each child. Item #1 asks about sleep duration; children sleeping <9 h per night were categorized as ‘abnormal’, and those sleeping $\geq$9 h were categorized as ‘normal’ [26]. Item #2 asks about time required to fall asleep; children taking > 30 min to fall asleep were categorized as ‘abnormal’, and those requiring $\leq$ 30 min were categorized as ‘normal’ [27]. Items #3–26 use the same 5-point Likert scale; responses of 3–5 (‘1 or 2 times a week’ or more often) were categorized as ‘abnormal’, whereas responses of 1 (‘never’) or 2 (‘rarely/1 or 2 times a month or less’) were categorized as ‘normal’.

2.2.3. Nighttime pain measure

We administered a questionnaire on sleep quality and sleep-related characteristics, which we created as part of the larger study. This questionnaire included an item which asked caregivers to report the frequency of their child's experience of nighttime pain on a 5-point scale similar to that of the SDSC. We categorized children as having significant nighttime pain if it occurred at least one night per week; this factor was used in our univariate and multivariate analyses.

2.2.4. Child characteristics

We collected additional data from medical charts and the Quebec CP Registry [28] including: 1) demographic information (sex and age), 2) details about the child's medical condition, including CP subtype and degree of motor impairment based on the Gross Motor Function Classification System (GMFCS) level [29], and 3) details about comorbidities, including auditory, visual and cognitive impairment and verbal functioning. For our analyses, we classified children's ages based on the categories used by the PedsQL Modules [toddler (3–4 years), young child (5–7 years) and child (8–12 years)], and dichotomized GMFCS levels as I–III (ambulatory) vs. IV/V (non-ambulatory), CP subtype as spastic quadriplegia/dyskinesia vs. other, moderate/severe cognitive impairment vs. none/mild impairment, and nonverbal vs. verbal functioning. For the children with available medical charts (n = 100), we collected information about whether the child had epilepsy and whether it was controlled (child on antiepileptic medication with no seizures in the last year) or active (child on antiepileptic medication with $\geq$1 seizure in the last year). To account for the many comorbidities that were present in children in our sample in our multivariate models, we created a dichotomous variable (‘significant comorbidity’) to indicate if a child had one or more of the following conditions: visual, auditory, cognitive impairment and/or epilepsy.

Study data were inputted and managed using Research Electronic Data Capture (REDCap) [30], a web-based application designed to support data capture for research studies, hosted at the Research Institute of the McGill University Health Centre.

2.3. Statistical analyses

Descriptive statistics were tabulated using means and SDs for continuous variables and proportions for categorical variables. Univariate and multiple logistic regressions were conducted to examine the association between predictor variables and impaired Total, Physical and Psychosocial HRQoL; odds ratios (ORs) and 95% confidence intervals (CIs) were estimated. In the multivariate analyses, we adjusted for clinically-relevant and statistically significant predictor variables: sleep problems, nighttime pain, age group and non-ambulatory status and the presence of one or more significant comorbidity. We performed the Hosmer–Lemeshow goodness of fit tests to assess our multivariate statistical models.

To further assess the relationship between specific sleep problems and Psychosocial HRQoL, we conducted univariate analyses to test the association between sleep problems on each of the 26 items on the SDSC and impaired HRQoL on the Psychosocial subscales (Emotional, Social and School Functioning) and determined the ORs (95% CIs). Finally, we evaluated the relationship between the PedsQL CP Module subscale scores (Daily Activities, School Activities, Movement and Balance, Pain and Hurt, Fatigue, Eating Activities, Speech and Communication) and sleep problems (total and DIMS T-scores on the SDSC) by generating scatter plots and Pearson correlation coefficients by ambulatory status (ie, GMFCS level I–III vs. IV/V). $P < 0.05$ was considered statistically significant. Bonferroni corrections were applied to account for multiple comparisons associated with each set of analyses. Statistical analyses were performed using IBM SPSS Statistics 24.0 for Windows (IBM Corp., Armonk, N.Y., USA).

3. Results

Overall, 146 children with CP were included in our study. The characteristics of these children are presented in Table 1. The mean age of children was 6.9 ± 2.9; 24.7% (36/146) were non-ambulatory (GMFCS levels IV/V) and 32.9% (48/146) were of the spastic quadriplegic or dyskinetic CP subtypes. Questionnaires were completed by mothers in 81.5% of cases (119/146). The overall response rate for the larger study was 41.7% (150/360); child characteristics (sex, age and CP subtype) did not vary between the children of caregivers who participated and did not participate in the study.

3.1. HRQoL on the PedsQL Generic Core Module

Table 2 presents HRQoL values for our sample of children with CP, as reported by caregivers on the PedsQL Generic Core Module, including mean scores and the number of children 1 SD and 2 SDs below the population mean for the Total score, summary scores and subscale scores. Approximately one third of children with CP had impaired Total HRQoL (ie, were more than 2 SDs below the population mean for the PedsQL Total score); twice as many children had impaired Physical compared to Psychosocial Health scores. Among the Psychosocial Health subscales, the greatest number of children were impaired on the Emotional Functioning subscale, followed by the Social Functioning and School Functioning subscales.

3.2. Child-related factors associated with HRQoL

Tables 3a, 3b and 3c show the results of the univariate and multiple logistic regressions between child-related factors and abnormal HRQoL scores (Total, Physical Summary, and Psychosocial Summary scores, respectively). Covariates in all three regression models included young child age (5–7 years), non-ambulatory status (GMFCS levels IV/V), presence of nighttime pain ($\geq$1 night per week), significant comorbidity (presence of severe visual,

Table 1
Characteristics of our study sample of 146 children with cerebral palsy.

Characteristics		n	%
Sex	Male	89	61.0
Age group	Toddler (3–4 years)	49	33.6
	Young child (5–7 years)	43	29.5
	Child (8–12 years)	54	37.0
GMFCS level	I	65	44.5
	II	40	27.4
	III	5	3.4
	IV	15	10.3
	V	21	14.4
CP subtype	Spastic-quadriplegic	32	21.9
	Spastic-hemiplegic	50	34.2
	Spastic-diplegic	35	24.0
	Spastic-other	11	7.5
	Dyskinetic	16	11.0
	Ataxic-hypotonic	2	1.4
Prematurity	Moderate-late preterm	23	15.8
	Very/extremely preterm	37	25.3
Cortical blindness/primary visual impairment		9	6.2
Severe auditory impairment		7	4.8
Moderate/severe cognitive impairment		24	16.4
Non-verbal		28	19.2
Gavage-fed		14	9.6
Epilepsy[a]	Controlled	15	15.0
	Active	17	17.0
Nighttime pain[b]		20	13.7
Sleep problems		30	20.5

[a] Information available only for a subset of 100 children with hospital medical records available for review.
[b] Nighttime pain reported by the caregivers of 138 children.

Table 2
Pediatric Quality of Life Inventory Generic Core Module scores for our sample of children with cerebral palsy and prevalence of children below population normative values.

PedsQL Generic Core Module Scales	N	Mean (SD)	Score more than 1 SD below population mean[a] n (%)	Score more than 2 SDs below population mean[a] n (%)
Total score	146	57.1 (17.9)	100 (68.5%)	49 (33.6%)
Physical Health Summary score	146	51.5 (27.8)	93 (63.7%)	56 (38.4%)
Psychosocial Health Summary score	142	61.0 (15.4)	89 (62.7%)	25 (17.6%)
Emotional Functioning subscale	145	65.0 (19.0)	60 (41.4%)	27 (18.6%)
Social Functioning subscale	142	59.5 (20.4)	89 (62.7%)	22 (15.5%)
School Functioning subscale[b]	132	58.4 (19.3)	71 (53.8%)	12 (9.1%)

PedsQL = Pediatric Quality of Life Inventory.
[a] Normative population mean in healthy children in the study by Varni et al., 2003.
[b] Children attending school or daycare only.

Table 3a
Univariate and multiple logistic regressions between child-related factors and an abnormal Total score on the Pediatric Quality of Life Inventory.

		Total score	Total score
		Crude OR (95% CI)	Adjusted OR (95% CI)
Age group	Toddler (3–4 years)	0.3 (0.1–0.6)**	–
	Young child (5–7 years)	2.2 (1.1–4.6)*	2.6 (0.9–7.7)
	Child (8–12 years)	1.5 (0.7–2.9)	–
Gender	Male	1.3 (0.6–2.7)	–
GMFCS level	IV/V	23.9 (8.8–65.5)**	30.1 (8.2–110.4)***
CP subtype	Quadriplegia/dyskinesia	8.1 (3.7–17.7)**	–
Prematurity (very/extreme preterm)		0.8 (0.4–1.8)	–
Cortical blindness/primary visual impairment		7.9 (1.6–39.7)*	–
Severe auditory impairment		5.4 (1.0–28.9)*	–
Moderate/severe cognitive impairment		6.5 (2.5–16.9)**	–
Non-verbal		9.6 (3.7–25.1)**	–
Gavage-fed		9.1 (2.4–34.3)**	–
Epilepsy		4.5 (1.8–11.2)**	–
Significant comorbidity		4.7 (2.2–9.8)**	3.3 (1.2–9.2)*
Nighttime pain		6.8 (2.4–19.4)**	4.0 (1.0–16.2)
Sleep problems		5.0 (2.1–11.6)**	3.8 (1.1–12.5)*

Crude OR: $^{*}p < 0.05$; $^{**}p < 0.003$ (Bonferroni correction for 16 comparisons). Adjusted OR: $^{*}p < 0.05$, $^{***}p < 0.001$.

Table 3b
Univariate and multiple logistic regressions between child-related factors and an abnormal Physical Health summary score on the Pediatric Quality of Life Inventory.

		Physical Health summary score Crude OR (95% CI)	Physical Health summary score Adjusted OR (95% CI)
Age group	Toddler (3–4 years)	0.5 (0.3–1.1)	–
	Young child (5–7 years)	2.4 (1.2–5.1)*	2.5 (0.9–6.4)
	Child (8–12 years)	0.8 (0.4–1.6)	–
Gender	Male	1.1 (0.6–2.2)	–
GMFCS level	IV/V	68.0 (15.2–305.0)**	58.3 (11.9–284–5)***
CP subtype	Quadriplegia/dyskinesia	11.7 (5.2–26.5)**	–
Prematurity (very/extreme preterm)		1.0 (0.5–2.1)	–
Cortical blindness/primary visual impairment		6.3 (1.3–31.4)*	–
Severe auditory impairment		10.7 (1.3–91.2)*	–
Moderate/severe cognitive impairment		5.9 (2.2–15.6)**	–
Non-verbal		11.8 (4.2–33.8)**	–
Gavage-fed		26.9 (3.4–212.4)**	–
Epilepsy		1.6 (0.7–3.8)	–
Significant comorbidity		2.1 (1.1–4.3)*	1.4 (0.4–5.7)
Nighttime pain		202.6 (1.0–6.7)	0.8 (0.4–5.7)
Sleep problems		1.8 (0.8–4.1)	1.1 (0.3–3.6)

Crude OR: $^{*}p < 0.05$; $^{**}p < 0.003$ (Bonferroni correction for 16 comparisons). Adjusted OR: $^{*}p < 0.05$, $^{***}p < 0.001$.

Table 3c
Univariate and multiple logistic regressions between child-related factors and an abnormal Psychosocial Health summary score on the Pediatric Quality of Life Inventory.

		Psychosocial Health summary score Crude OR (95% CI)	Psychosocial Health summary score Adjusted OR (95% CI)
Age group	Toddler (3–4 years)	0.2 (0.1–0.8)*	–
	Young child (5–7 years)	2.7 (1.1–6.5)*	2.8 (1.0–7.6)
	Child (8–12 years)	1.2 (0.5–2.9)	–
Gender	Male	1.4 (0.6–3.6)	–
GMFCS level	IV/V	1.7 (0.7–4.5)	1.4 (0.4–4.3)
CP subtype	Quadriplegia/dyskinesia	1.3 (0.5–3.1)	–
Prematurity (very/extreme preterm)		0.5 (0.2–1.6)	–
Cortical blindness/primary visual impairment		0.8 (0.1–6.7)	–
Severe auditory impairment		0.8 (0.1–6.7)	–
Moderate/severe cognitive impairment		0.8 (0.2–2.8)	–
Non-verbal		0.8 (0.3–2.6)	–
Gavage-fed		1.5 (0.4–5.7)	–
Epilepsy		4.5 (1.5–13.5)*	–
Significant comorbidity		2.4 (1.0–5.9)*	1.0 (0.2–3.7)
Nighttime pain		3.2 (1.1–9.2)*	2.3 (0.8–6.3)
Sleep problems		8.7 (3.3–22.6)**	7.5 (2.5–22.5)

Crude OR: $^{*}p < 0.05$; $^{**}p < 0.003$ (Bonferroni correction for 16 comparisons). Adjusted OR: $^{*}p < 0.05$.

auditory, cognitive impairment and/or epilepsy) and sleep problems (abnormal total score on the SDSC). The Hosmer–Lemeshow goodness of fit test for each logistic regression model confirmed that models were appropriate [PedsQL Total score, X2(df = 5): 2.881, p = 0.718, Physical Health summary score, X2(df = 6): 2.550, p = 0.863 and Psychosocial Health summary score, X2(df = 6): 4.569, p = 0.660].

3.2.1. Total HRQoL

As seen in Table 3a, on univariate analysis, nine child-related factors were found to be associated with an increased odds of impaired HRQoL (abnormal PedsQL Total score), including CP phenotype, several comorbidities, nighttime pain and sleep problems. On multivariate analysis, non-ambulatory status (adjusted OR 30.1, 95% CI 8.2–110.4), sleep problems (adjusted OR 3.8, 95% CI 1.1–12.5) and significant comorbidity (adjusted OR 3.3, 95% CI 1.2–9.2) were associated with impaired HRQoL, adjusted for young child age (5–7 years) and nighttime pain.

3.2.2. Physical HRQoL

Table 3b shows that factors associated specifically with an abnormal Physical Health summary score were a quadriplegic/dyskinetic CP subtype, non-ambulatory, cognitive impairment, non-verbal and gavage-fed. Non-ambulatory children with CP had approximately 58 times the odds of having an abnormal Physical Health summary score (adjusted OR 58.3, 95% CI 11.9–284.5).

3.2.3. Psychosocial HRQoL

Table 3c shows that, among the factors analyzed, sleep problems alone were significantly associated with an abnormal Psychosocial Health summary score. Children with sleep problems had more than seven times the odds of having an abnormal Psychosocial Health summary score (adjusted OR 7.5, 95% CI 2.5–22.5).

3.3. Specific sleep problems associated with psychosocial HRQoL

To further examine the relationship between Psychosocial HRQoL and sleep problems, we conducted univariate analyses to produce ORs (95% CIs) for abnormal Emotional, Social and School Functioning scores (subscales of Psychosocial HRQoL) by specific sleep problems on the SDSC. Only the Emotional Functioning subscale showed significant associations with specific sleep problems; the following six sleep problems were associated with an increased odds of having an abnormal Emotional Functioning subscale score (from highest to lowest ORs): having nightmares which are not remembered the next day (OR 17.0, 95% CI 4.1–70.3), feeling anxious or afraid when falling asleep (OR 12.8, 95% CI 4.9–34.0), sweating excessively while falling asleep (OR 7.4, 95% CI 2.9–19.2),

Table 4a
Association between Pediatric Quality of Life Cerebral Palsy Module subscale scores and total scores on the Sleep Disturbance Scale for Children for ambulatory and non-ambulatory children with cerebral palsy.

PedsQL CP Module Scales	SDSC Total score All children	SCDC Total score GMFCS I–III	SDSC Total score GMFCS IV/V
Daily activities (n = 146)	−0.11	0.01	0.10
School activities (n = 97)	−0.15	−0.10	−0.08
Movement and balance (n = 146)	−0.27**	−0.22*	−0.12
Pain and hurt (n = 142)	−0.48**	−0.43**	−0.56**
Fatigue (n = 144)	−0.27**	−0.23**	−0.24
Eating activities (n = 141)	−0.20**	−0.10	−0.01
Speech and communication (n = 96)	−0.15	−0.13	0.09

PedsQL = Pediatric Quality of Life Inventory; SDSC = Sleep Disturbance Scale for Children. Pearson's correlation coefficients; *$p < 0.05$, **$p < 0.01$.

experiencing vivid dream-like scenes while falling asleep (OR 6.4, 95% CI 2.5–15.9), difficulty getting to sleep at night (OR 6.3, 95% CI 2.4–16.1), and going to bed reluctantly (OR 4.5, 95% CI 1.8–11.0).

3.4. *Relationship between CP-specific HRQoL and sleep by ambulatory/non-ambulatory status*

Tables 4a and 4b and Figs. 1a and 1b show the association between scores on each of the seven PedsQL CP Module subscales and the total scores and DIMS scores on the SDSC for ambulatory and non-ambulatory children in our sample. Significant moderate negative correlations were found between the Pain and Hurt subscale scores and total scores on the SDSC, with a stronger correlation in non-ambulatory children. Significant weak negative correlations (ie, coefficients of correlation from −0.3 to −0.1) were found between the Movement and Balance and Fatigue subscales and total score on the SDSC in ambulatory children with CP exclusively. DIMS scores showed a significant association with the Pain and Hurt subscale scores, with a moderate negative association and a weak negative association in non-ambulatory and ambulatory children, respectively. School Activities subscale scores showed a significant weak-moderate association with DIMS scores in non-ambulatory children exclusively.

4. Discussion

In our study of Canadian preschool- and school-aged children with a wide range of CP severities and comorbidities, we found that one in three children have an overall HRQoL that is more than 2 standard deviations below the average value reported for a large population of typically developing children. We found that children with CP who had impaired HRQoL on the PedsQL were 30 times more likely to be non-ambulatory, four times more likely to have sleep problems and three times more likely to have significant comorbidity, when controlling for the effects of young age group (5–7 years) and nighttime pain (one night/per week or more often). Among these factors, non-ambulatory status alone was associated with 58 times the odds of having PedsQL Physical summary scores that were beyond the 2 SD cut-off; sleep problems alone were associated with more than seven times the odds of having PedsQL Psychosocial summary scores that were beyond the 2 SD cut-off. Several sleep problems on the SDSC were associated with an increased odds of impaired emotional functioning, specifically. Among these six sleep problems, half were symptoms of DIMS, namely reluctance going to bed, anxiety or fear when falling asleep and difficulty getting to sleep. The remaining sleep problems were reflective of symptoms of behavioural insomnia (bedtime resistance and difficult falling asleep), nightmares, excessive sweating and vivid dream-like scenes when falling asleep. Impaired HRQoL due to pain was associated with more sleep problems on the SDSC; this association was more pronounced in non-ambulatory compared with ambulatory children.

Our study is one of the few to consider sleep problems as a potentially important child characteristic associated with HRQoL in children with CP. To our knowledge, we are the first to report a significant role for sleep problems in the overall HRQoL of children with CP when controlling for the effects of other known modifiers of HRQoL, such as level of motor functioning and pain. We found a particularly strong association between sleep problems and psychosocial HRQoL in our sample of children with CP and found that specific symptoms of DIMS and unrestful sleep were associated with impaired HRQoL related to the emotional functioning dimension of psychosocial HRQoL. Sandella and colleagues [19] found a similar association between caregiver-reported symptoms of insomnia on the Pediatric Sleep Questionnaire and psychosocial HRQoL on the PedsQL, controlling for the effects of GMFCS, in a small sample (n = 41) of school-aged, higher functioning (~12% GMFCS IV/V) children with CP. Our study included preschool-aged children and a wide range of CP phenotypes, including a significant proportion of non-ambulatory children (~25%), two groups of children with CP who are not always well represented in neurodevelopmental disorder research. By including a diverse sample, we were able to compare the effect of sleep problems on CP-specific HRQoL outcomes by ambulatory status; we found, for example, that sleep problems have a greater negative impact on pain-related HRQoL in non-ambulatory children than in ambulatory children with CP. Recruitment of families into our study relied in part on the Quebec Cerebral Palsy Registry, a

Table 4b
Association between Pediatric Quality of Life Cerebral Palsy Module subscale scores and disorders of initiation and maintenance of sleep scores on the Sleep Disturbance Scale for Children for ambulatory and non-ambulatory children with cerebral palsy.

PedsQL CP Module Scales	DIMS score All children	DIMS score GMFCS I–III	DIMS score GMFCS IV/V
Daily activities (n = 146)	−0.15*	0.03	−0.08
School activities (n = 97)	−0.19*	0.03	−0.33*
Movement and balance (n = 146)	−0.21**	−0.05	−0.13
Pain and hurt (n = 142)	−0.33**	−0.19*	−0.45**
Fatigue (n = 144)	−0.13	−0.07	−0.04
Eating activities (n = 141)	−0.23**	0.02	−0.16
Speech and communication (n = 96)	−0.22*	−0.09	−0.15

PedsQL = Pediatric Quality of Life Inventory; DIMS = Disorder of initiation and maintenance of sleep. Pearson's correlation coefficients; *$p < 0.05$, **$p < 0.01$.

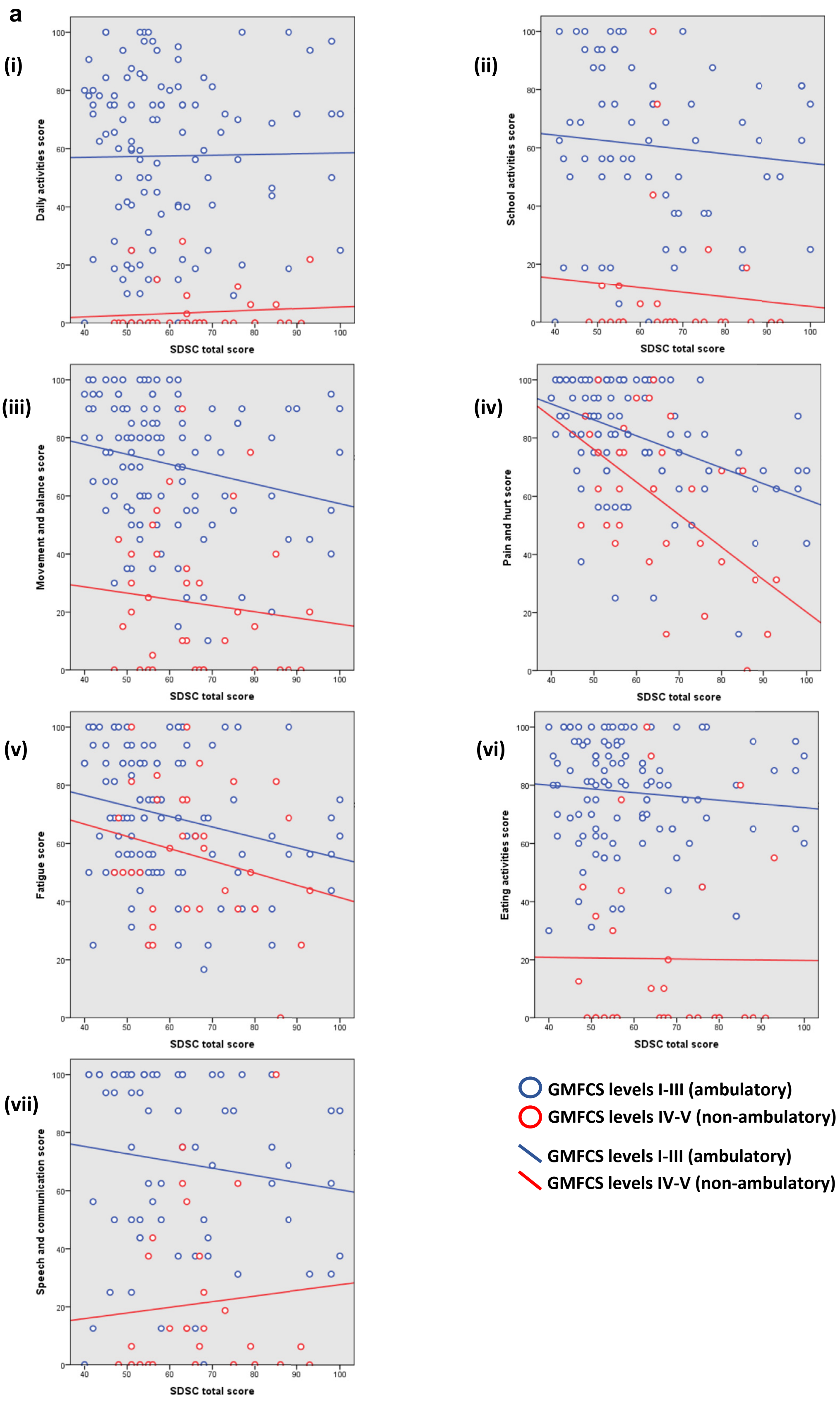

Fig. 1a. Relationship between total scores on the Sleep Disturbance Scale for Children (SDSC) and scores on the Pediatric Quality of Life Inventory Cerebral Palsy Module scales for ambulatory and non-ambulatory children. (i) Daily Activities , (ii) School Activities , (iii) Movement and Balance , (iv) Pain and Hurt, (v) Fatigue, (vi) Eating Activities, (vii) Speech and Communication. Correlation coefficients and significance values shown in Table 4a.

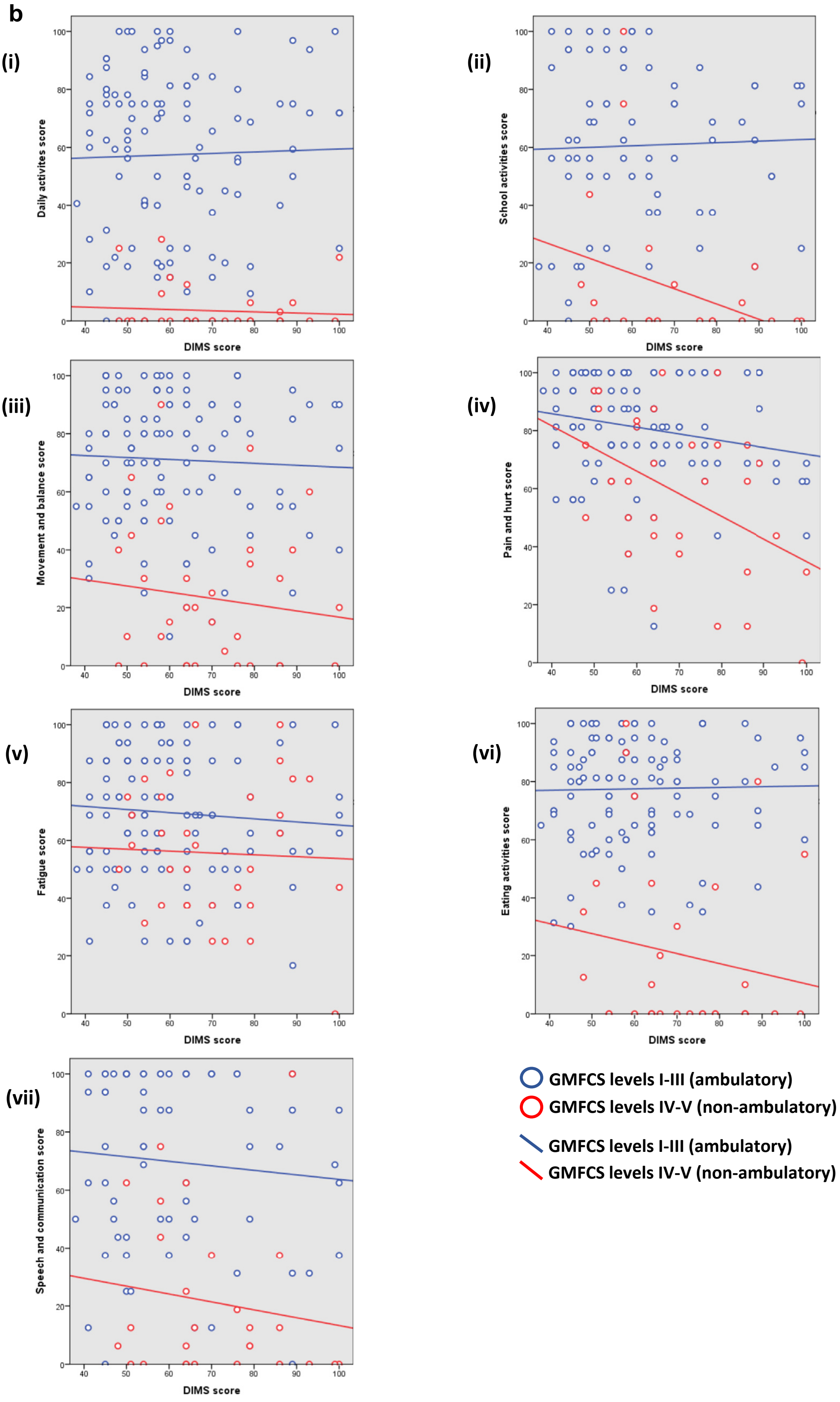

Fig. 1b. Relationship between disorders of initiation and maintenance of sleep (DIMS) scores on the Sleep Disturbance Scale for Children (SDSC) and scores on the Pediatric Quality of Life Inventory Cerebral Palsy Module scales for ambulatory and non-ambulatory children. (i) Daily Activities , (ii) School Activities , (iii) Movement and Balance, (iv) Pain and Hurt, (v) Fatigue, (vi) Eating Activities , (vii) Speech and Communication. Correlation coefficients and significance values shown in Table 4b.

valuable source of data for population-based research on CP. This approach allowed us to include families living in rural areas adjacent to the Greater Montreal Area and further enhances the generalizability of our study findings.

We acknowledge that there are limitations inherent to our study. First, our study relies on proxy-reported measures of HRQoL for the children in our sample. Although the self-report version of the PedsQL has been used in other studies of children with CP, it can only be used in children five years and older and in children with sufficient cognitive capabilities to provide responses to the questions. We therefore used the caregiver-report versions of the PedsQL to allow the greatest inclusion of children aged 3–4 years in our study, as well as older children with physical and/or cognitive limitations that would have prevented them from reporting on their own HRQoL. Previous studies have demonstrated that child and caregiver reports of various aspects of HRQoL show good correspondence [15,31]. While our study relied on caregiver-proxy assessments of perceived child HRQoL, we did not evaluate caregivers' psychological state, which has been proposed to mediate the responses given by caregivers regarding their children's HRQoL [32]. Our study also does not account for other socio-familial and/or environmental factors that could affect HRQoL as well as sleep problems. We used a cross-sectional design, and our study is thus limited in explaining the direct cause(s) of impaired HRQoL in children with CP. Although our data suggest a possible decrease in HRQoL in children with CP aged 5–7 years compared with their younger and older peers, our design also precludes us from examining more closely the possible changes in HRQoL that may occur as children progress through the various phases of development.

5. Conclusion

Our results suggest that sleep, non-ambulatory status and presence of comorbidities are important determinants of HRQoL in children with CP. Sleep problems may lead to decreased overall HRQoL, with a particularly negative impact on psychosocial HRQoL. Moreover, sleep problems may be amenable to intervention, and are thus another potentially modifiable factor that, once identified and treated, can lead to significant improvements in the HRQoL of children with CP. Appropriate and regular inquiry about sleep problems, difficulties with emotional functioning as well as pain by members of children's medical-rehabilitation team will encourage the implementation of initiatives to promote these aspects of children's health and optimize the HRQoL of the child with CP and their family.

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Conflicts of interest

None.

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