Research in Developmental Disabilities 95 (2019) 103500

Contents lists available at ScienceDirect

Research in Developmental Disabilities

journal homepage: www.elsevier.com/locate/redevdis

Behavioral difficulties, sleep problems, and nighttime pain in children with cerebral palsy☆

Linda Horwood[a,b], Patricia Li[a,b], Elise Mok[b], Maryam Oskoui[a,b,c], Michael Shevell[a,b,c], Evelyn Constantin[a,b,*]

[a] Department of Pediatrics, Montreal Children's Hospital, McGill University Health Centre, Montreal, Quebec, Canada
[b] Research Institute, McGill University Health Centre, Montreal, Quebec, Canada
[c] Department of Neurology/Neurosurgery, Montreal Children's Hospital, McGill University Health Centre, Montreal, Quebec, Canada

ARTICLE INFO

Number of reviews completed is 2

Keywords:
Behavior
Cerebral palsy
Sleep
Pain
Strengths and Difficulties Questionnaire

ABSTRACT

Background: Children with cerebral palsy (CP) may be at risk of behavioral difficulties.
Aims: 1) Determine the prevalence of behavioral difficulties in preschool- and school-aged children with CP and 2) Assess the association between behavioral difficulties and a) sleep problems, b) nighttime pain and c) child characteristics (age, CP phenotype, comorbidities).
Methods and procedures: Caregivers of 113 children with CP aged 4–12 years [mean (SD) age = 7.4 (2.5) years; 61.9% male] completed the Strengths and Difficulties Questionnaire, Sleep Disturbance Scale for Children and a sleep quality questionnaire to assess child behavior, sleep and nighttime pain, respectively.
Outcomes and results: 25.6% of children (17.6% preschool-aged; 29.1% school-aged) had behavioral difficulties. Sleep problems (odds ratio [OR] 9.1, 95% confidence interval [CI] 3.4–24.4) and nighttime pain (OR 4.1, 95% CI 1.5–11.5) were associated with behavioral difficulties. Sleep problems remained significantly associated with behavioral difficulties (adjusted OR 7.5, 95% CI 2.6–21.4) when adjusted for nighttime pain, age and non-ambulatory status.
Conclusions and implications: Behavioral difficulties were reported in one in four children with CP and were associated with sleep problems and nighttime pain. Identifying and treating behavioral difficulties, sleep problems or nighttime pain is important in the care of children with CP.

What this paper adds?

In this cross-sectional study of prospectively recruited Canadian children with cerebral palsy (CP) aged 4–12 years, we report that 25% of children with CP had behavioral difficulties on the Strengths and Difficulties Questionnaire, with higher rates in school-aged compared to preschool-aged children. Rates of behavioral difficulties in preschool-aged children with CP were double those previously reported for typically-developing children (~20% vs. 10%). In children with CP, sleep problems and nighttime pain increased the likelihood of behavioral problems by 9- and 4-fold, respectively. Behavioral difficulties were associated mainly with sleep problems relating to disorders of initiation and maintenance of sleep (DIMS). To our knowledge, this is the first study to consider sleep problems and nighttime pain as child-related factors associated with behavioral difficulties in children with CP; sleep problems were

☆ The paper is part of a special issue on Sleep in Disabilities.

* Corresponding author at: Montreal Children's Hospital / McGill University Health Centre, 1001 Decarie Boulevard, AS1 2335, Montreal, Quebec, H4A 3J1, Canada.
E-mail address: evelyn.constantin@mcgill.ca (E. Constantin).

https://doi.org/10.1016/j.ridd.2019.103500
Received 26 January 2019; Received in revised form 26 August 2019; Accepted 27 September 2019
Available online 17 October 2019
0891-4222/

the most significant factor associated with behavioral difficulties in our sample of children with CP. Other novel features include examination of the association between individual sleep problems and behavioral difficulties; sleep problems relating specifically to DIMS were associated with a greater likelihood of behavioral difficulties. The study supports an important interplay between behavioral difficulties, sleep problems and pain, a notable gap in the literature identified by a recent systematic review on the co-occurrence of impairments in children with CP. The findings highlight that clinicians and other members of the health care team should consider the co-occurrence of behavior problems, sleep issues, and pain in the management of children with CP.

1. Introduction

The nature and extent of behavioral difficulties experienced by children with cerebral palsy (CP), the most common physical impairment in children (Oskoui, Coutinho, Dykeman, Jette, & Pringsheim, 2013), has received increasing attention over the last decade. A recent position statement from the Canadian Paediatric Society provided recommendations for the assessment and management of mental health symptoms in children with neuromotor disabilities, including CP (Klein et al., 2016). Behavior problems, in addition to the primary motor impairment and potential presence of other comorbidities that are common in CP, such as cognitive impairment and epilepsy, can further compound a child's limitations in activities of daily living and social participation, and lead to an even greater reduction in the quality of life of the child and their family (Butcher, Wind, & Bouma, 2008; Lowes, Clark, & Noritz, 2016; Majnemer, Shevell, Rosenbaum, Law, & Poulin, 2007; Raina et al., 2005; Romeo et al., 2010; Tseng et al., 2016).

Behavioral difficulties, including hyperactivity, difficulties in executive functioning, and emotional dysregulation, are among the many negative consequences of poor sleep quality in otherwise healthy children (Beebe, 2011; Sadeh, Gruber, & Raviv, 2002). Sleep problems are now recognized as a common comorbidity in children with CP (Novak, Hines, Goldsmith, & Barclay, 2012), and yet, the potential association between sleep disturbances and the behavioral functioning of children with CP has received limited attention. One study by Romeo and colleagues (Romeo et al., 2014) examined multiple variables, including behavior problems on the Child Behavior Checklist, as potentially being associated with sleep problems. However, no studies have looked at sleep problems and their impact on behavior. To date, studies have examined the influence of other important CP-related comorbidities on behavior, and reported that behavioral difficulties were more frequent in *school-aged* children with CP who had more significant functional limitations (Parkes et al., 2008), greater cognitive impairment (Parkes et al., 2008), severe pain (Parkes et al., 2008), and epilepsy (Carlsson, Olsson, Hagberg, & Beckung, 2008).

The Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997) provides a simple, reliable and validated method of screening for important areas of potential behavioral difficulties in typically-developing children (Goodman, 2001). Whereas the SDQ has been used in studies of school-aged children (Brossard-Racine et al., 2012; Parkes et al., 2008) and adolescents (Brossard-Racine et al., 2013) with CP, to our knowledge, no studies have utilized this simple screening tool to report on the prevalence and types of behavioral difficulties found in *preschool-aged* children with CP. Our study aimed to 1) describe the prevalence and types of behavioral difficulties using the SDQ in children with CP overall and by age, in *both* preschool-aged (4–5 years) and school-aged (6–12 years) children, 2) assess the association between behavioral difficulties and a) sleep problems, b) nighttime pain, and c) child characteristics (age, CP phenotype, comorbidities) and 3) identify specific sleep problems that are associated with an increased risk of behavioral difficulties. We hypothesized that children with CP who had caregiver-reported sleep problems and nighttime pain would be at greater risk of behavioral difficulties (high/very high SDQ total difficulties scores) compared with their peers with CP who did not have significant sleep problems and nighttime pain.

2. Methods

2.1. Participants and procedures

As part of a broader cross-sectional study investigating the prevalence of sleep disorders in children with CP (Horwood et al., 2018), 150 children were prospectively recruited from 2013 to 2016 in hospital neurology clinics and from a regional CP registry (Horwood et al., 2018). Inclusion criteria for the larger study were: age 3–12 years; diagnosis of CP by a neurologist; living at home with a primary caregiver or guardian able to read and complete the questionnaires. For the current study, we excluded children < 4 years of age, as the SDQ version utilized was designated for use in children 4 years of age and older. The study protocol was approved by the hospital's Research Ethics Board and participating caregivers provided written informed consent to the research and publication of results.

2.2. Behavioral difficulties

Behavioral difficulties were reported by caregivers on the parent-report version of the SDQ (Goodman, 1997), a 25-item behavioral screening tool with acceptable reliability and validity in typically-developing children (Goodman, 2001). The SDQ assesses five domains of behavioral difficulties: emotional symptoms, conduct problems, hyperactivity, peer problems, and prosocial behaviors (not considered in this study); no item assesses for behavioral sleep problems specifically. As per the scoring guidelines for the SDQ (SDQ Information for researchers & professionals about the Strengths & Difficulties Questionnaire, 2016), we accounted for missing data by prorating based on the mean of non-missing items of the relevant domain score (when at least 3 of 5 items were completed). Domain scores and the total difficulties score (sum of the four domains) were classified based on cut-off points determined from the distribution of scores in a large normative sample [80% ('normal'), 10% ('borderline'), 5% ('high') and 5% ('very high')] (Goodman,

2001). For our analyses, 'high'/'very high' scores and 'normal'/'borderline' scores were categorized as 'abnormal' and 'normal', respectively.

2.3. Sleep problems

The Sleep Disturbance Scale for Children (SDSC), a validated screening tool for pediatric sleep disorders, asks caregivers to report on the frequency of 26 common sleep issues found in children (Bruni et al., 1996). Responses to each of the 26 items are given on a 5-point Likert scale; the sum of items provides a total score, which can then be classified as normal (T-score $\leq$70) or abnormal (T-scores > 70). The questionnaire also provides six subscores for specific sleep disorders, such as disorders of initiation and maintenance of sleep and sleep breathing disorders, however, subscores can be normal while the total score can be abnormal. Thus, to account for children experiencing overall clinically concerning sleep issues, we included an abnormal total score on the SDSC as a predictor variable for behavioral difficulties in our univariate logistic regression analyses. To further detail the relationship between sleep problems and behavioral difficulties, we dichotomized responses on each of the 26 items on the SDSC to classify whether the sleep issue was a problem for each child; we then examined for an association between each sleep issue and behavioral difficulties. Item #1 asks about sleep duration; responses were dichotomized such that children sleeping < 9 h per night were categorized as 'abnormal', and those sleeping $\geq$9 h were categorized as 'normal', as all children in our study should have been getting more than 9 h of sleep according to current guidelines (Hirshkowitz et al., 2015) ('9-11 h' was the longest sleep duration that caregivers could endorse on the SDSC). 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' (Dement & Guilleminault, 2016). 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'.

2.4. Nighttime pain

We administered an additional questionnaire on sleep quality and sleep-related characteristics, which we created as part of the larger study (Horwood et al., 2018). This questionnaire included an item which asked: "Does your child experience any kind of pain at night?"; caregivers indicated the frequency of their child's nighttime pain on a 5-point Likert scale similar to that of the SDSC. We categorized children as having significant pain if caregivers reported that pain occurred at least one night per week.

2.5. Child characteristics

We collected additional data from medical charts and the CP Registry including: 1) demographic information (sex and age), 2) details about the child's medical condition, including CP subtype and degree of motor impairment, i.e., Gross Motor Function Classification System (GMFCS) level (Palisano et al., 1997), and 3) details about comorbidities, including cognitive impairment and verbal functioning. For our analyses, we classified children as preschool- (4–5 years) vs. school-aged (6–12 years), and dichotomized GMFCS as levels 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 non-verbal vs. verbal functioning. For children with medical charts (n = 73), we collected information about confirmed or suspected neurodevelopmental/ neuropsychiatric disorders (i.e., autism spectrum disorder, attention-deficit/hyperactivity disorder, anxiety, and other behavioral disorders), and epilepsy.

Study data were inputted and managed using Research Electronic Data Capture (REDCap) (Harris et al., 2009), a web-based application designed to support data capture for research studies, hosted at our institution.

2.6. Statistical analyses

Descriptive statistics were tabulated using means and standard deviations (SD) for continuous variables and proportions for categorical variables. Chi-square tests were used to test the distributions of total difficulties scores on the SDQ in our sample against expected (population-based) proportions of 80%, 10%, 5% and 5% for 'normal', 'borderline', 'high' and 'very high' categories, respectively. Univariate logistic regressions were conducted to examine the association between child-related factors and abnormal SDQ domain and total difficulties scores. Odds ratio (ORs) and 95% confidence interval (CIs) were estimated. Multivariate logistic regression was subsequently performed with an abnormal total difficulties score as the outcome variable. In the multivariate logistic regression analysis, we adjusted for clinically relevant variables (school-aged, non-ambulatory status) (Parkes et al., 2008), in addition to the presence of sleep problems and nighttime pain. The Hosmer-Lemeshow goodness of fit test was performed to assess this multivariate statistical model.

We conducted a subgroup analysis of children with medical records available (n = 73) to examine the possible association between behavior difficulties and other neurodevelopmental/neuropsychiatric disorders (yes/suspected vs. none) and epilepsy (yes vs. no). We repeated, for the abnormal total difficulties score only, the univariate logistic regression analyses and multivariate logistic regression analysis (described above) for this subset of children, this time including neurodevelopmental/neuropsychiatric disorders and epilepsy as factors.

Finally, to assess the relationship between specific sleep issues and behavioral difficulties, we conducted univariate logistic regression analyses to test the association between individual sleep problems (i.e., an 'abnormal' status on each of the 26 items on the SDSC, as described above) and an abnormal total difficulties score and reported the OR (95% CI). $P < 0.05$ was considered

Table 1
Demographic and child characteristics of 113 children with cerebral palsy.

Characteristics		n	% of total
Sex	*Male*	70	61.9%
Age group	*Preschool-aged (4–5 years)*	34	30.1%
	School-aged (6–12 years)	79	69.9%
GMFCS	*I*	55	48.7%
	II	27	23.9%
	III	3	2.7%
	IV	14	12.4%
	V	14	12.4%
CP subtype	*Spastic-quadriplegic*	23	20.4%
	Spastic-hemiplegic	43	38.1%
	Spastic-diplegic	26	23.0%
	Spastic-other	5	4.4%
	Dyskinetic	14	12.4%
	Ataxic-hypotonic	2	1.8%
Cognitive impairment	*Moderate/severe*	19	16.8%
	None/mild	82	72.6%
	Uncertain	12	10.6%
Non-verbal communication		22	19.5%
Sleep problems[†]		26	23.0%
Nighttime pain[††]		19	16.8%
Neurodevelopmental/neuropsychiatric disorder*		27	37.0%
Epilepsy*		25	34.2%

[†] Abnormal total score on the Sleep Disturbance Scale for Children.
[††] Nighttime pain once a week or more often.
* Information available for a subset of 73 children with medical charts.

statistically significant. All statistical analyses were performed using IBM SPSS Statistics 24.0 for Windows (IBM Corp., Armonk, N.Y., USA).

3. Results

Of the 150 children in our larger study, 113 children met our inclusion criteria; we excluded children < 4 years (n = 30) and children with incomplete SDQ data (n = 7). The demographic characteristics of our study sample are detailed in Table 1. Mean (SD) age was 7.4 (2.5) years, 84 (74.3%) children were ambulatory (GMFCS levels I-III) and 37 (32.7%) children had a spastic quadriplegic or dyskinetic CP subtype.

The majority of caregivers participating in the study were mothers (93, 82.3%). More children were recruited from neurology clinics (69, 61.1%) than the CP registry (44, 38.9%). The overall response rate for our study was 39.9% and varied by recruitment approach (89.1% in clinic vs. 28.9% by mail). The characteristics (sex, age and CP subtype) of the children of non-respondent caregivers and participating caregivers were compared; these characteristics did not differ between the two groups.

3.1. Behavioral difficulties on the SDQ

Table 2 shows the distribution of the four domain and total difficulties scores on the SDQ; the distribution of the total difficulties scores observed for our sample of children with CP differed significantly from the expected population-based distribution of scores in typically-developing children (X^2, $p < 0.1 \times 10^{-6}$). A post-hoc power calculation based on the known population prevalence of children scoring in the clinically abnormal range for total difficulties scores (high/very high; 10%) and our sample prevalence of 25.6% in 113 children determined that our study had a post-hoc power of > 80% based on an alpha of 0.05. Across all children (4–12

Table 2
Prevalence (n, %) of behavioral difficulties on the Strengths and Difficulties Questionnaire in 113 children with cerebral palsy.

SDQ Domains	Normal	Borderline	High	Very High
Observed				
Emotional Symptoms	70 (61.9%)	12 (10.6%)	16 (14.2%)	15 (13.3%)
Conduct Problems	74 (65.5%)	21 (18.6%)	16 (14.2%)	2 (1.8%)
Hyperactivity	64 (56.6%)	24 (21.2%)	11 (9.7%)	14 (12.4%)
Peer Problems	64 (56.6%)	17 (15.0%)	9 (8.0%)	23 (20.4%)
Total Difficulties	**62 (54.9%)**	**22 (19.5%)**	**12 (10.6%)**	**17 (15.0%)**
Expected				
Domain and Total Difficulties	80%	10%	5%	5%

SDQ = Strengths and Difficulties Questionnaire.

Table 3
Univariate and multiple logistic regressions for behavioral difficulties on the Strengths and Difficulties Questionnaire by child-related factors in children with cerebral palsy.

Characteristics	N	Emotional Symptoms Crude OR (95% CI)	Conduct Problems Crude OR (95% CI)	Hyperactivity Crude OR (95% CI)	Peer Problems Crude OR (95% CI)	Total Difficulties Crude OR (95% CI)	Total Difficulties Adjusted OR (95% CI)
Male	70	1.2 (0.5–2.7)	1.0 (0.3–2.7)	1.8(0.7–4.7)	0.9(0.4–2.0)	**1.2 (0.5 –3.0)**	–
School-aged	79	2.2 (0.8–5.9)	2.4(0.7–9.0)	1.1 (0.4–3.0)	1.4(0.6–3.6)	**1.9 (0.7–5.2)**	**1.1(0.3 –3.5)**
Non-ambulatory	29	0.5 (0.2–1.4)	0.8(0.3–2.8)	0.7(0.2–2.1)	1.3(0.5–3.2)	**0.7 (0.3–2.0)**	**0.5(0.1 –1.8)**
Quadriplegic/ Dyskinetic	37	0.6 (0.3–1.6)	0.8(0.2–2.3)	0.6(0.2–1.6)	1.1(0.5–2.6)	**0.9(0.4 –2.2)**	–
Cognitive impairment	19	0.3 (0.1–1.2)	0.9(0.2–3.6)	0.3 (0.1–1.6)	0.6(0.2–2.1)	**0.5(0.1 –1.8)**	–
Non-verbal	22	0.4 (0.1–1.3)	0.5(0.1–2.2)	0.7 (0.2–2.4)	0.9(0.3–2.7)	**0.6(0.2 –1.9)**	–
†Sleep problems	26	6.1*** (2.3–15.6)	4.6** (1.6–13.3)	3.8** (1.5–10.0)	2.3(0.9–5.8)	**9.1*** (3.4–24.4)**	**7.5***(2.6 –21.4)**
††Nighttime Pain	19	4.7** (1.7–13.4)	3.0(0.9–9.3)	1.2(0.4–3.8)	1.5(0.5–4.2)	**4.1** (1.5–11.5)**	**2.7 (0.8–9.1)**

*p < 0.05.
** p < 0.01.
*** p < 0.001.
† Abnormal total score on the Sleep Disturbance Scale for Children; ††Nighttime pain once a week or more often.

years), the prevalence of children scoring in the clinically abnormal range for the SDQ domains was as follows: peer problems were the most common behavioral difficulty (28.4%), followed by emotional symptoms (27.5%), hyperactivity (22.1%) and conduct problems (16.0%). The prevalence of behavioral problems was determined for preschool-aged (4–5 years) and school-aged (6–12 years) children by SDQ domain; the prevalence of scores in the clinically abnormal range were: emotional symptoms in 17.6% and 31.6%, conduct problems in 8.8% and 19.0%, hyperactivity in 20.6% and 22.8%, and peer problems in 23.5% and 30.4% of preschool- and school-aged children, respectively. Abnormal total difficulties scores were found in 17.6% and 29.1% of preschool- and school-aged children, respectively.

3.2. Child characteristics and sleep problems associated with behavioral difficulties

Table 3 presents the results of our univariate logistic regression analyses; nighttime pain and sleep problems (an abnormal total score on the SDSC) were the only two factors examined which were significantly associated with behavioral problems in children with CP. Specifically, sleep problems were associated with emotional symptoms, conduct problems, hyperactivity and an abnormal total difficulties score; pain was associated with emotional symptoms and an abnormal total difficulties score Multivariate logistic regression (Table 3) demonstrated that children with sleep problems had more than seven times greater odds of having an abnormal total difficulties score (adjusted OR 7.5, 95% CI 2.6–21.4), adjusted for nighttime pain, school-age and non-ambulatory status. The Hosmer-Lemeshow goodness of fit test confirmed that the model was appropriate [X^2 (df = 4): 0.53, p = 0.97].

3.3. Epilepsy and neurodevelopmental/neuropsychiatric disorders: association with behavioral difficulties

For 73 children (64.6%) in our sample, hospital medical records were available and information about current epilepsy and neurodevelopmental/neuropsychiatric disorders was collected.

Twenty-five children (34.2%) had epilepsy and 27 children (37.0%) had diagnosed or suspected neurodevelopmental/neuropsychiatric disorders: 12.3% (9/73) had autism spectrum disorder, 27.4% (20/73) had attention deficit/hyperactivity disorder, 5.5% (4/73) had anxiety, 6.8% (5/73) had a behavioral disorder; 12.3% (9/73) had more than one of these disorders. An abnormal total difficulties score was present in 27.4% (20/73) of children. We conducted univariate logistic regression analyses as above with epilepsy and neurodevelopmental/neuropsychiatric disorders (all disorders, individual disorders and more than one disorder) as seven additional factors that may be associated with an abnormal total difficulties score. As with our previous analyses, pain (OR 10.0, 95% CI 2.3–43.6) and sleep problems (OR 22.8, 95% CI 5.8–89.7) were significantly associated with an abnormal total difficulties score, along with epilepsy (OR 4.6, 95% CI 1.5–13.8) and having more than one neurodevelopmental/neuropsychiatric disorder (OR 13.7, 9.5% CI 2.5–74.1). Multivariate logistic regression, adjusted for school-age, non-ambulatory status, nighttime pain, epilepsy and more than one neurodevelopmental/neuropsychiatric disorder showed that children with sleep problems had more than eight times the odds of having an abnormal total difficulties score (adjusted OR 8.4, 95% CI 1.6–45.5).

Table 4
Odds ratios (95% confidence intervals) for an abnormal total difficulties score on the Strengths and Difficulties Questionnaire by sleep problems on the Sleep Disturbance Scale for Children in 113 children with cerebral palsy.

Sleep problem (item #)	N	Total Difficulties OR (95% CI)
Sleeps less than 9 h (1)	39	1.0 (0.4–2.4)
Takes more than 30 min to fall asleep (2)	26	2.7 (1.1–6.9)*
Goes to bed reluctantly (3)	36	2.1 (0.9–5.1)
Has difficulty getting to sleep at night (4)	45	5.2 (2.1–13.1)***
Feels anxious or afraid when falling asleep (5)	25	6.2 (2.4–16.3)***
Startles or jerks parts of the body while falling asleep (6)	25	3.0 (1.2–7.7)
Shows repetitive actions while falling asleep (7)	12	0.9 (0.2–3.8)
Experiences vivid dream-like scenes while falling asleep (8)	22	5.0 (1.9–13.5)**
Sweats excessively while falling asleep (9)	23	3.9 (1.5–10.3)**
The child wakes up more than twice per night (10)	37	4.5 (1.8–10.9)**
After waking up in the night, has difficulty to fall asleep again (11)	28	4.5 (1.8–11.5)**
Has frequent twitching or jerking of legs/often changes position/kicks the covers off the bed (12)	51	2.5 (1.1–6.0)*
Has difficulty breathing in the night (13)	10	0.7 (0.1–3.5)
Gasps for breath or is unable to breathe during sleep (14)	3	1.5 (0.1–16.8)
Snores (15)	32	1.2 (0.5–3.0)
Sweats excessively during the night (16)	19	1.4 (0.5–4.2)
Has been observed sleepwalking by you (17)	3	–
Has been observed talking in their sleep by you (18)	22	2.5 (0.9–6.8)
Grinds their teeth during sleep (19)	31	4.5 (1.8–11.1)**
Wakes from sleep screaming or confused, has no memory of these events the next morning (20)	4	9.6 (1.0–96.1)
Has nightmares which they don't remember the next day (21)	9	7.4 (1.7–31.8)**
Is unusually difficult to wake up in the morning (22)	21	2.7 (1.0–7.3)
Awakes in the morning feeling tired (23)	39	4.9 (2.0–12.1)***
Feels unable to move when waking up in the morning (24)	6	3.2 (0.6–16.9)
Experiences daytime somnolence (25)	24	4.2 (1.6–10.9)**
Falls asleep suddenly in inappropriate situations (26)	3	6.1 (0.5–70.5)

* $p < 0.05$.
** $p < 0.01$.
*** $p < 0.001$.

3.4. Specific sleep problems associated with behavioral difficulties

Table 4 presents the results of the univariate logistic regression analysis of behavioral difficulties on the SDQ by specific sleep problems on the SDSC. The following twelve sleep problems were associated with an increased odds of having behavioral difficulties in children with CP (from highest to lowest ORs): nightmares, feeling anxious or afraid when falling asleep, difficulty getting to sleep at night, experiencing vivid dream-like scenes while falling asleep, waking up in the morning feeling tired, waking up more than twice per night, difficulty falling back to sleep after waking up in the night, grinding teeth during sleep (sleep bruxism), daytime somnolence, sweating excessively while falling asleep, taking more than 30 min to fall asleep and frequent twitching or jerking of legs/changing position often during sleep.

4. Discussion

One in four children with CP had behavioral difficulties based on caregiver-report on the SDQ, with the most commonly reported issues being peer problems and emotional symptoms. Sleep disturbances and nighttime pain were found to be significantly associated with increased odds of behavioral difficulties by approximately nine- and four-fold, respectively. Specific sleep problems relating mainly to symptoms of disorders of initiation and maintenance of sleep (e.g., difficulty falling asleep at night, frequent nighttime awakenings) were significantly associated with increased odds of behavioral difficulties by approximately three- to seven-fold.

The prevalence of behavioral difficulties in children with CP was just over 25%, which is markedly elevated compared to the general pediatric population, where one would expect to encounter behavioral difficulties in only *1 in 10* children (Goodman, 2001). Rates of behavioral difficulties in school-aged children in our study were similar to those reported by Brossard-Racine and colleagues (Brossard-Racine et al., 2012) in their sample of school-aged children with CP (i.e., 29.1% vs. 27.6%, respectively). Our rates of behavioral problems (domain and total scores) fall within the 95% CIs determined in a large population-based study of children aged 8–12 years (Parkes et al., 2008), with only hyperactivity (22.8%) falling outside their 95% CI of 29–33%. In the preschool-aged children with CP in our study, clinically abnormal behavioral problems, including emotional symptoms, hyperactivity, peer problems and overall behavioral difficulties were already present at about *twice* the expected prevalence of that found in typically-developing children (Goodman, 1997) (i.e., each ~20% vs. 10%). This finding suggests that even prior to formally entering the classroom environment, young children with CP have higher rates of behavioral difficulties than in their typically-developing peers. We further found that the frequency of emotional symptoms, conduct problems and overall difficulties were approximately double in school-aged compared to preschool-aged children with CP. Future directions may include exploring the importance of earlier screening for

behavioral problems in very young children with CP to provide earlier assessments and tools/treatment prior to entering school.

Our study supports an important interplay between sleep problems, behavioral difficulties and pain, a notable gap in the literature identified by Novak and colleagues (Novak et al., 2012) in their recent systematic review on the co-occurrence of impairments in children with CP. Previous studies have identified an association between sleep problems and pain in children with CP (Hemmingsson, Stenhammar, & Paulsson, 2009; Horwood et al., 2018; McCabe, Blackmore, Abbiss, Langdon, & Elliott, 2015) but these studies did not also examine the association of sleep problems and pain with behavioral difficulties. We found that nighttime pain was the second most important predictor (after sleep problems) of severe behavior problems on univariate logistic regression analysis. Children with CP who experienced nighttime pain at least one night per week had five times greater odds of emotional symptoms, and four times greater odds of behavioral difficulties overall. Although we found a strong association between behavioral difficulties and sleep problems, due to the inherent limitations of a cross-sectional study design, we are unable to determine directionality and causality. However, our multivariate logistic regression analysis found that, over and beyond the influence of the other factors assessed, children with significant sleep problems had more than seven times greater odds of experiencing clinically concerning behavioral difficulties.

Oberlander and colleagues (Oberlander, O'Donnell, & Montgomery, 1999) illustrate the highly complex and multidirectional relationship between sleep disturbances, pain and cognitive-behavioral functioning in their review on pain in children with CP, outlining the potential influences of the underlying (and often extensive) brain lesion, difficulties with cognitive functioning and/or communication, the severity and number of comorbid health conditions and related interventions, the child's developmental stage and their broader familial and socio-environmental surroundings. Lewin and Dahl (Lewin & Dahl, 1999) have proposed a model for the interaction between sleep, pain and behavior: pain can lead to reductions in sleep quantity and quality, either directly via nociceptive sensations that cause delayed sleep onset and sleep fragmentation, or indirectly, by inducing a state of arousal/anxiety, heightened vigilance to the onset of pain (particularly in chronic pain, as in CP), and other cognitive and affective disturbances, which in turn lead to poor sleep. Sleep deficiency has many cognitive-behavioral sequelae, including increased sleepiness, decreased motivation and compromised emotional regulation and attention which may decrease the ability to cope with pain and fuel a cycle of ongoing sleep disturbances and decreased cognitive-behavioral functioning (Lewin & Dahl, 1999).

In addition to sleep problems and pain, we found associations between epilepsy and neurodevelopmental/neuropsychiatric disorders and behavioral difficulties in our subgroup analysis. Children with CP who had epilepsy had nearly five times the odds of behavioral difficulties, a finding which is in agreement with a previous study which also used the SDQ and found that children with CP who had epilepsy had higher total difficulties scores compared to those without epilepsy (Carlsson et al., 2008). Children in our study who were identified as having more than one suspected or diagnosed neurodevelopmental/neuropsychiatric disorder had nearly 14 times the odds of high/very high behavioral difficulties scores on the SDQ. This finding is not surprising given that this tool assesses several domains of behavioral functioning, and children with more than one neurodevelopmental/neuropsychiatric disorder would foreseeably present with a greater array of behavioral difficulties. In our multivariate logistic regression analysis, controlling for these and other child-related factors, sleep problems were again the only significant predictor of behavioral difficulties in children with CP.

To our knowledge, our study is the first to examine the relationship between specific sleep problems and behavioral difficulties in children with CP. We found that several sleep problems increased the odds of behavioral difficulties by four- to six-fold: feeling anxious or afraid when falling asleep, difficulty getting to sleep, vivid dreaming, frequent awakenings, difficulty resuming sleep, sleep bruxism. These sleep problems could all be related to anxiety, or more specifically, sleep-related anxiety. Not unsurprisingly, waking in the morning feeling tired, a potential consequence of poor sleep, was also highly associated with behavioral difficulties. Disorders of initiation and maintenance of sleep are indeed the most commonly reported sleep disorder in studies which have used the SDSC in children with CP (Atmawidjaja, Wong, Yang, & Ong, 2014; Horwood et al., 2018; Newman, O'Regan, & Hensey, 2006; Romeo et al., 2014). Again, our design limits our ability to ascertain whether children with CP have high rates of sleep problems due to behavioral disturbances, such as emotional symptoms or hyperactivity, or whether the susceptibility of children with CP to sleep disturbances due to their underlying disease and/or comorbidities results in more adverse behavioral outcomes, such as difficulties with emotional regulation and attention.

Our study has limitations. First, our design was cross-sectional. Second, we did not include a concurrent control group with which to compare our prevalence estimates for behavioral difficulties in our sample of children with CP. However, normative population data are available for the SDQ based on a sample of close to 10,000 British 5–15-year old children (Goodman, 2001), and we statistically compared those expected prevalence estimates against those observed in our study. The study by Goodman unfortunately does not provide an age distribution for the sample, nor does it detail the prevalence of behavioral difficulties by age group. Third, we used caregiver-completed questionnaires to assess behavioral difficulties, sleep problems and pain in children with CP. Although the use of self-report questionnaires may not be ideal, a reliance on caregiver reports in this and other studies of children with neurodevelopmental disabilities allows for the inclusion of children across the spectrum of ages and severity of disabilities, including those with severe cognitive delay, communication difficulties and/or fine motor impairments. We chose our primary outcome measure, the SDQ (Goodman, 2001), specifically due to its prior validation for use in children with CP (Parkes et al., 2008). Our study also did not include an objective assessment of physiological sleep disorders, such as sleep-disordered breathing or restless leg syndrome, which may have yielded a greater prevalence of children with CP with these sleep disorders than that determined by caregiver questionnaires alone. Of interest, the prevalence of snoring in our sample was high (32/113 = 28.3%). This is in keeping with other studies that have reported a high risk of sleep-disordered breathing in children with CP, particularly those who are GMFCS IV and V (Dawson et al., 2013; Hill, Parker, Allen, Paul, & Padoa, 2009). Finally, child behavior and associated child characteristics were the focus of the current study, and thus, we did not collect information on caregiver factors that might have shown important mediating

associations with child behavioral difficulties, such as demographic variables, caregiver mental health, parenting approaches, stress, coping strategies and access to resources. Furthermore, though we found elevated rates of behavior problems in our sample, it is possible that some caregivers of children with neurodevelopmental disabilities may not readily recognize these disturbances in their children or may consider them to be "part of the disability" and not modifiable/amenable to treatment. The collection of more extensive information about and from caregivers, as well as adding behavioral assessments from other informants (such as teachers), would more clearly delineate the nature, extent and context (e.g., home vs. classroom environment) of children's behavioral difficulties and the perceived impact on the primary caregiver and other family members. To date, the consensus among studies of caregivers is that behavior problems in children with CP confer an additional burden beyond that of the child's objective impairments, including increased parental stress and depression, and decreased quality of life for the family (Butcher et al., 2008; Lowes et al., 2016; Majnemer, Shevell, Rosenbaum, Law, & Poulin, 2007; Raina et al., 2005; Romeo et al., 2010; Tseng et al., 2016).

In conclusion, our study found that one in four children with CP has clinically significant behavioral difficulties involving a level of emotional disturbance and/or social impairment that is likely to warrant intervention. Our study supports a strong link between behavioral difficulties and sleep problems, as well as a potential association between pain and behavioral difficulties in children with CP, a population known to have elevated rates of behavioral difficulties, sleep problems, and chronic pain. Our findings indicate that clinicians and other members of the health care team should consider the co-occurrence of behavioral problems, pain and sleep issues in the management of children with CP.

Declaration of Competing Interest

None.

Acknowledgments

The authors would like to thank the administrative staff affiliated with the clinics at the Montreal Children's Hospital (Isilda Nascimento and Melissa Acciaioli) and the coordinators at the Quebec Cerebral Palsy Registry (Sasha Dyck Holzinger, Christopher Saunders and Lise Doré) for their assistance with facilitating the recruitment of families. The authors also thank the families for their interest and willingness to participate in this study.

Dr. Horwood's doctoral work on the study was funded in part by awards from the Fonds de recherche du Québec-Santé (FRQ-S), the Kids Brain Health Network (KBHN), the Research Institute of the McGill University Health Care Centre (RI-MUHC) and McGill University. Dr. Li holds grants from the FRQ-S, the RI-MUHC and the Canadian Institutes of Health Research (CIHR). Dr. Oskoui holds grants from the FRQ-S, CIHR, the Cerebral Palsy Alliance Foundation, and KBHN. Dr. Shevell holds grants from the KBHN. Dr. Constantin holds grants from the KBHN, CIHR and RI-MUHC. The RI-MUHC is supported in part by the FRQ-S. The Cerebral Palsy Registry is funded by the FRQ-S and the KBHN.

References

Atmawidjaja, R. W., Wong, S. W., Yang, W. W., & Ong, L. C. (2014). Sleep disturbances in Malaysian children with cerebral palsy. *Developmental Medicine and Child Neurology, 56*, 681–685.

Beebe, D. W. (2011). Cognitive, behavioral, and functional consequences of inadequate sleep in children and adolescents. *Pediatric Clinics of North America, 58*(3), 649–665.

Brossard-Racine, M., Hall, N., Majnemer, A., Shevell, M. I., Law, M., Poulin, C., et al. (2012). Behavioural problems in school age children with cerebral palsy. *European Journal of Paediatric Neurology, 16*(1), 35–41. https://doi.org/10.1016/j.ejpn.2011.10.001.

Brossard-Racine, M., Waknin, J., Shikako-Thomas, K., Shevell, M., Poulin, C., Lach, L., ... Majnemer, A. (2013). Behavioral difficulties in adolescents with cerebral palsy. *Journal of Child Neurology, 28*(1), 27–33. https://doi.org/10.1177/0883073812461942.

Bruni, O., Ottaviano, S., Guidetti, V., Romoli, M., Innocenzi, M., Cortesi, F., et al. (1996). The Sleep Disturbance Scale for Children (SDSC). Construction and validation of an instrument to evaluate sleep disturbances in childhood and adolescence. *Journal of Sleep Research, 5*(4), 251–261.

Butcher, P. R., Wind, T., & Bouma, A. (2008). Parenting stress in mothers and fathers of a child with a hemiparesis: Sources of stress, intervening factors and long-term expressions of stress. *CCH Child: Care, Health and Development, 34*(4), 530–541.

Carlsson, M., Olsson, I., Hagberg, G., & Beckung, E. (2008). Behaviour in children with cerebral palsy with and without epilepsy. *Developmental Medicine and Child Neurology, 50*(10), 784–789.

Dawson, N. C., Padoa, K. A., Bucks, R. S., Allen, P., Evans, H., McCaughey, E., et al. (2013). Ventilatory function in children with severe motor disorders using night-time postural equipment. *Developmental Medicine and Child Neurology, 55*, 751–757. https://doi.org/10.1111/dmcn.12149.

Dement, W. C., & Guilleminault, C. (2016). Sleep Disorders: The State of the Art. *Hospital Practice Hospital Practice, 8*(11), 57–71.

Goodman, R. (1997). The Strengths and Difficulties Questionnaire: a research note. *Journal of Child Psychology and Psychiatry, 38*(5), 581–586.

Goodman, R. (2001). Psychometric properties of the strengths and difficulties questionnaire. *Journal of the American Academy of Child and Adolescent Psychiatry, 40*(11), 1337–1345.

Harris, P. A., Taylor, R., Thielke, R., Payne, J., Gonzalez, N., & Conde, J. G. (2009). Research electronic data capture (REDCap)—A metadata-driven methodology and workflow process for providing translational research informatics support. *Journal of Biomedical Informatics, 42*(2), 377–381.

Hemmingsson, H., Stenhammar, A. M., & Paulsson, K. (2009). Sleep problems and the need for parental night-time attention in children with physical disabilities. *Child: Care, Health and Development, 35*, 89–95.

Hill, C. M., Parker, R. C., Allen, P., Paul, A., & Padoa, K. A. (2009). Sleep quality and respiratory function in children with severe cerebral palsy using night-time postural equipment: A pilot study. *Acta Paediatrica, 98*, 1809–1814.

Hirshkowitz, M., Whiton, K., Albert, S. M., Alessi, C., Bruni, O., DonCarlos, L., ... Ware, J. C. (2015). National Sleep Foundation's updated sleep duration recommendations: final report. *Sleep Health, 1*(4), 233–243. https://doi.org/10.1016/j.sleh.2015.10.004.

Horwood, L., Mok, E., Li, P., Oskoui, M., Shevell, M., & Constantin, E. (2018). Prevalence of sleep problems and sleep-related characteristics in preschool- and school-aged children with cerebral palsy. *Sleep Medicine, 50*, 1–6.

Klein, Benjamin, Canadian Paediatric Society, & Mental Health and Developmental Disabilities Committee (2016). Mental health problems in children with neuromotor disabilities. *Paediatrics & Child Health, 21*(2), 93–96. https://doi.org/10.1093/pch/21.2.93.

Lewin, D. S., & Dahl, R. E. (1999). Importance of sleep in the management of pediatric pain. *Journal of Developmental and Behavioral Pediatrics: JDBP, 20*(4), 244–252.
Lowes, L., Clark, T. S., & Noritz, G. (2016). Factors associated with caregiver experience in families with a child with cerebral palsy. *Journal of Pediatric Rehabilitation Medicine, 9*(1), 65–72.
Majnemer, A., Shevell, M., Rosenbaum, P., Law, M., & Poulin, C. (2007). Determinants of life quality in school-age children with cerebral palsy. *Journal of Pediatrics, 151*(5), 470–475 475.e471–473.
McCabe, S. M., Blackmore, A. M., Abbiss, C. R., Langdon, K., & Elliott, C. (2015). Sleep concerns in children and young people with cerebral palsy in their home setting. *Journal of Paediatrics and Child Health, 51*(12), 1188–1194.
Newman, C. J., O'Regan, M., & Hensey, O. (2006). Sleep disorders in children with cerebral palsy. *Developmental Medicine and Child Neurology, 48*, 564–568.
Novak, I., Hines, M., Goldsmith, S., & Barclay, R. (2012). Clinical prognostic messages from a systematic review on cerebral palsy. *Pediatrics, 130*, e1285–1312.
Oberlander, T. F., O'Donnell, M. E., & Montgomery, C. J. (1999). Pain in children with significant neurological impairment. *Journal of Developmental and Behavioral Pediatrics: JDBP, 20*(4), 235–243.
Oskoui, M., Coutinho, F., Dykeman, J., Jette, N., & Pringsheim, T. (2013). An update on the prevalence of cerebral palsy: A systematic review and meta-analysis. *Developmental Medicine and Child Neurology, 55*(6), 509–519.
Palisano, R., Rosenbaum, P., Walter, S., Russell, D., Wood, E., & Galuppi, B. (1997). Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Developmental Medicine and Child Neurology, 39*(4), 214–223.
Parkes, J., White-Koning, M., Dickinson, H. O., Thyen, U., Arnaud, C., Beckung, E., ... Colver, A. (2008). Psychological problems in children with cerebral palsy: a cross-sectional European study. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 49*(4), 405–413. https://doi.org/10.1111/j.1469-7610.2007.01845.x.
Raina, P., O'Donnell, M., Rosenbaum, P., Brehaut, J., Walter, S. D., Russell, D., ... Wood, E. (2005). The health and well-being of caregivers of children with cerebral palsy. *Pediatrics, 115*(6), E626–E636. https://doi.org/10.1542/peds.2004-1689.
Romeo, D. M., Cioni, M., Distefano, A., Battaglia, L. R., Costanzo, L., Ricci, D., ... Mercuri, E. (2010). Quality of life in parents of children with cerebral palsy: is it influenced by the child's behaviour? *Neuropediatrics, 41*(3), 121–126. https://doi.org/10.1055/s-0030-1262841.
Romeo, D. M., Brogna, C., Quintiliani, M., Baranello, G., Pagliano, E., Casalino, T., ... Mercuri, E. (2014). Sleep disorders in children with cerebral palsy: neurodevelopmental and behavioral correlates. *Sleep Medicine, 15*, 213–218. https://doi.org/10.1016/j.sleep.2013.08.793.
Sadeh, A., Gruber, R., & Raviv, A. (2002). Sleep, neurobehavioral functioning, and behavior problems in school-age children. *Child Development, 73*(2), 405–417.
SDQ Information for researchers and professionals about the Strengths and Difficulties Questionnaire (2016). *SDQ Information for researchers and professionals about the Strengths and Difficulties Questionnaire.* www.sdqinfo.com.
Tseng, M. H., Chen, K. L., Shieh, J. Y., Lu, L., Huang, C. Y., & Simeonsson, R. J. (2016). Child characteristics, caregiver characteristics, and environmental factors affecting the quality of life of caregivers of children with cerebral palsy. *Disability and Rehabilitation, 38*(24–26), 2374–2382.