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A systematic review and meta-analysis of the prevalence of sleep problems in children with cerebral palsy: how do children with cerebral palsy differ from each other and from typically developing children?

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ABSTRACT

Background: Up to 85% of children with neurodevelopmental disorders have sleep problems, compared with 25% of typically developing children. Children with cerebral palsy (CP)may have risk factors (brain injury, physical disability, and comorbidities) that make them more likely to have sleep problems compared with typically developing children.
Objective: To determine prevalence of sleep problems in children with CP.
Methods: We conducted a systematic review and meta-analysis to report on the prevalence of sleep problems in children with CP, within subgroups (age, CP phenotype, presence of impairments [auditory, visual, and cognitive], and presence of epilepsy) and compared with control groups of healthy children. We searched eight relevant electronic databases from their respective start dates until September 2018.
Results: 23 full-text articles (n=2,908 children with CP) were included in the review. All studies were cross-sectional and examined caregiver-reported sleep measures. The Sleep Disturbance Scale for Children (SDSC) was the most commonly used questionnaire. No study met all Joanna Briggs Institute quality assessment criteria for prevalence studies; selection, coverage, classification, and/or confounding biases were present in all studies. Using a random effects model with a Freeman-Tukey double arcsine transformation, the pooled prevalence was 23.4% (95% confidence interval [CI] 18.8-28.4%; n=9 studies) for an abnormal total score on the SDSC and 26.9% (95% CI 21.5-32.7%; n=9 studies) for disorders of initiation and maintenance of sleep, the most prevalent sleep problem reported. For the studies that reported prevalence for control groups of healthy children (n=4 studies), sleep problems were generally more prevalent in the CP group.
Conclusion: The prevalence of sleep problems in children with CP is high. There is notable variability in the prevalence of sleep problems between subgroups of children with CP. Future studies using questionnaires validated in children with CP and objective measures (such as polysomnography or actigraphy) in well-described, large, broadly recruited samples are recommended.

Introduction

Sleep disturbances can have broad consequences on children potentially negatively affecting growth[1] and development[2] as well as behavior and cognition.[3,4] The impact of childhood sleep problems may be far-reaching and also have a significant influence on the psychological and physical functioning of the child's family. In children with complex medical and/or behavioral needs such as those with neurodevelopmental disorders (NDDs), sleep problems may contribute to further reduction in quality of life for both children[5,6] and their caregivers.[7]

The literature to date has reported rates of sleep problems in heterogeneous groups of children with NDDs (i.e., combining children with autism, attention deficit hyperactivity disorder, fetal alcohol syndrome, and others), suggesting rates that may be as high as 85%[8]

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which is much higher than the prevalence of sleep problems in typically developing children, which is reported to be about 20-30%.[9] A NDD that should be of particular interest in sleep medicine is cerebral palsy (CP).

With a prevalence of about 2 in 1,000 children,[10] CP, a NDD of movement and posture resulting from a lesion arising in the brain during early fetal or infant development, is the most common cause of childhood physical disability worldwide.[11] The phenotype of children with CP can vary widely depending on the location and extent of damage to the brain. The Gross Motor Function Classification System (GMFCS) is a commonly used tool to describe the degree of ambulation of children with CP.[12] Children who are levels I-III are considered ambulatory (ambulate independently) and individuals who are levels IV-V are nonambulatory (do not ambulate independently) and are considered a more severe CP phenotype. Children with CP also vary in terms of the extent of the body that is affected. Hemiplegia, a milder phenotype of CP, primarily affects one upper limb, whereas quadriplegia, a more severe phenotype, causes motor deficits in all four limbs and other areas of the body.

In addition to the motor difficulties that characterize CP, children with CP may have one or more comorbidities which may markedly increase the likelihood of sleep problems.[13] Epilepsy, a common comorbidity of CP, may cause nocturnal seizures that awaken the child.[14–16] Significant visual impairments, which are also common in CP, may delay sleep onset because light perception may be reduced to the point that the child's endogenous circadian rhythm is disrupted.[17,18] There have also been some studies demonstrating that children with CP may have a dysfunctional release of hormones that contribute to circadian regulation.[19,20] In children with developmental delay (such as those children with CP), a few studies have shown abnormal secretion of melatonin, a hormone that plays a role in the generation of sleep.[19] Pain from increased muscle tone or spasms or discomfort from orthotic devices may cause awakenings.[21,22] Despite frequently having these and other significant risk factors for sleep problems, children with CP have received less attention in sleep research than children with other NDDs such as autism spectrum disorder and attention deficit hyperactivity disorder.

Children with CP are believed to have a spectrum of sleep problems similar to typically developing children. The most common sleep problem in both typically developing children and children with CP is insomnia, defined as "a repeated difficulty with sleep initiation, duration, consolidation, or quality that occurs despite age-appropriate time and opportunity for sleep and results in daytime functional impairment for the child and/or family".[23] Another common sleep problem is sleep-disordered breathing (SDB) which refers to a spectrum of sleep-related breathing disorders (from primary snoring as the mildest disturbance to obstructive sleep apnea as the most severe condition).[24] Disorders of arousal (parasomnias) are characterized by incomplete arousals from deep sleep that manifest as a broad variety of emotional and motor behaviors, including sleepwalking, sleep terrors, and arousals from sleep in a heightened state of confusion.[25] Sleep-wake transition disorders involve parasomnias that occur either during the transition from wakefulness to sleep, or between sleep stages, and may manifest as abnormal behaviors. The most common sleep-wake transition disorder is sleep talking (somniloquy); other disorders include rhythmic movement disorder, nocturnal leg cramps, and myoclonus (brief involuntary muscle twitches).[26] Excessive somnolence, or sleepiness, can be the result of several physiological and psychological conditions. These include narcolepsy, SDB, anxiety, and depression.[27] Sleep hyperhidrosis involves excessive sweating while falling asleep or during sleep and may or may not be associated with excessive daytime sweating.[28]

In children with CP, who often have a number of comorbidities in addition to difficulties with movement, sleep problems such as insomnia may exacerbate these impairments and further reduce quality of life. A comprehensive description and synthesis of the prevalence and types of sleep problems reported in children with CP may raise the awareness of health professionals working with children with CP, supporting timely screening and intervention for sleep problems in this pediatric population. Lelis et al [13] performed an integrative review which focussed on identifying and synthesizing the literature on the nature of sleep disorders and related factors in children with CP.[13] Though this review by Lelis et al provided important insight on the nature of sleep disorders in children with CP, this review did not provide data on the prevalence of sleep problems in children with CP. To our knowledge, no studies have synthesized existing literature to inform on the prevalence of sleep problems in children with CP. We conducted a systematic review and meta-analysis of the literature to determine the prevalence of sleep problems reported in children with CP, including prevalence within subgroups (age, CP phenotype, presence of impairments [auditory, visual, and cognitive], and presence of epilepsy) and compared with prevalence of sleep problems in healthy control children.

Methods

We registered the details of our systematic review with the International Prospective Register of Systematic Reviews (PROSPERO; identification number CRD42014004051).[29]

Search strategy

We developed a comprehensive search strategy in collaboration with an experienced medical librarian to identify relevant studies. We searched the following electronic databases: Allied and Complementary Medicine Database (AMED), BIOSIS Previews, Cochrane, Embase, Global Health, MEDLINE/PubMed, and Web of Science. We used terms related to sleep, cerebral palsy, and child, from the respective start dates of the databases through September 2018. Database-specific search terms are presented in Appendix 1. The reference lists of articles being considered at the full-text stage (see below) as well as review articles on the topic of sleep and NDDs were hand-searched for additional references appearing relevant to our systematic review.

Selection criteria

We considered for inclusion full-text articles published in English or French that included participants who were children (i.e., aged $\leq$18 years) with a diagnosis of CP where the prevalence of one or more sleep problems assessed by any tool was reported. To minimize selection bias, we excluded case reports and case series, as well as studies which used samples of children institutionalized in long-term care facilities, as these children were not considered to be representative of the overall population of children with CP. In cases where there were mixed samples of children (i.e., studies that included adults and/or other individuals with conditions other than CP), we contacted authors by email to obtain the required subgroup data; articles were excluded if specific sleep outcome data for children with CP were not available.

Selection process

A reviewer (L.H.) screened the titles and abstracts of retrieved studies using the predetermined selection criteria outlined previously. When articles could not be excluded based on the title and abstract, the full-text article was reviewed. The selection process from among potentially eligible articles was carried out by two independent reviewers (L.H. and E.C.). Disagreements regarding an article's inclusion were discussed, and agreements were reached by consensus.

Data extraction

For studies that met the selection criteria, a reviewer (L.H.) used a data collection form to extract the following information: authors, country of publication, study design, methods of recruitment, study setting, sample demographics (number, age, and gender of participants), participant characteristics, sleep assessment tool used, type of sleep problems assessed, prevalence rates of sleep problems overall, and in any subgroup(s) studied. For each included article, all extracted data were verified by a second author during the quality assessment process. Participant characteristics were decided a priori based on the literature and included age, frequencies of CP subtypes and GMFCS levels, and comorbidities that included epilepsy and auditory, visual, and cognitive impairments.

Quality assessment

We used the Joanna Briggs Institute (JBI) Critical Appraisal Checklist for Studies Reporting Prevalence Data[30,31] to guide our assessment of the quality of the included studies. The JBI checklist comprises 10 items. We divided item 9 into two separate questions to assess whether studies (a) identified and (b) accounted for important demographic and child-related factors. Each study was assessed by two independent authors and discrepancies in the assessments were resolved by consensus.

Data analyses

We performed a meta-analysis of studies that used the same sleep assessment tool using a random effects model approach with a Freeman-Tukey double arcsine transformation to ascertain overall pooled prevalence by sleep problem. We used metaprop, a statistical program to perform meta-analyses of proportions in Stata;[32] metaprop calculates heterogeneity within groups of >2 studies using the I^2 statistic.[33] I^2 statistics with p-values >0.01 were considered to have insignificant heterogeneity.[34] Meta-analyses were performed using Stata, version 15.1 (Stata Statistical Software: Release 15; StataCorp. 2017., StataCorp LLC, College Station, TX).

Results

Our search yielded 5,338 unique records. Figure 1 outlines the study selection process.[35] Twenty-three full-text articles,[5–7,36–55] including nonoverlapping samples of 2,908 children with CP and 593 healthy control children, met the inclusion criteria for our review. Details about the included studies are presented in Table 1. The years of publication ranged from 2000 to 2018, with 18 studies[5–7,36–40,42,43,45–47,49,50,52–54] having been published from 2010-2018. Studies were conducted in all of the following major continents: Africa (n=2),[38,50] Asia (n=4),[7,36,43,48] Australia (n=2),[44,45]

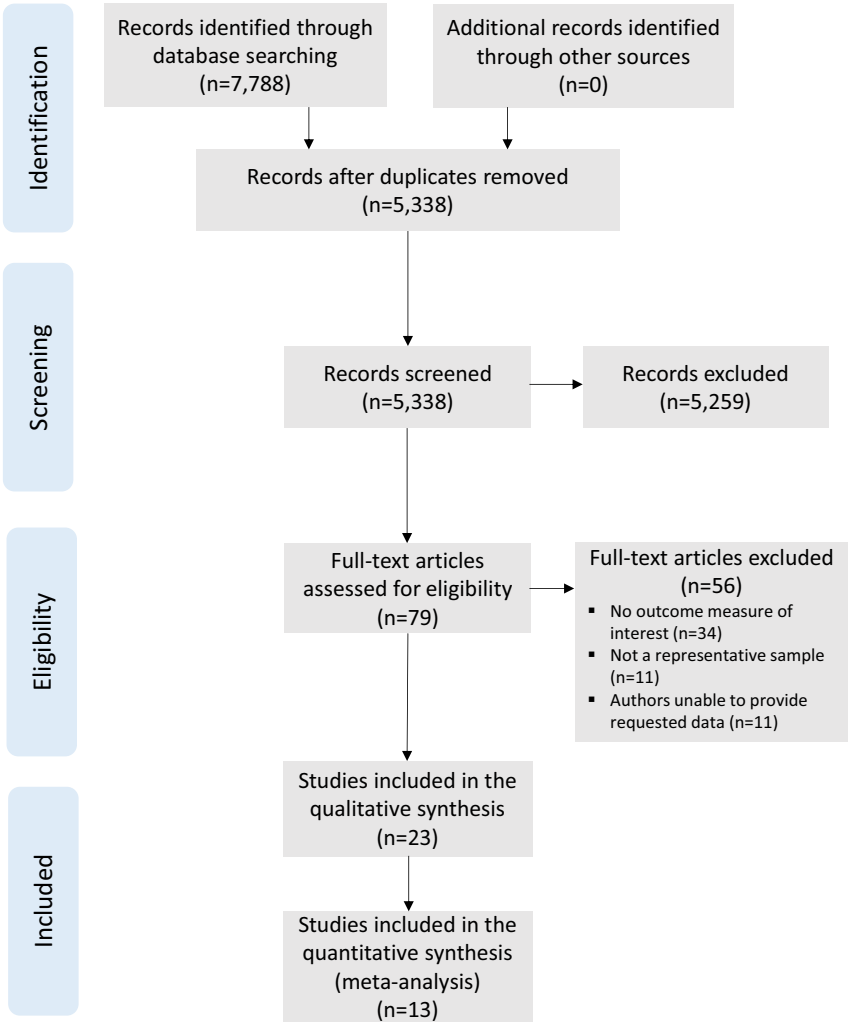


Fig. 1. Four-phase PRISMA flow diagram showing the number of studies identified, screened, eligible, and included in the systematic review and meta-analysis.

Table 1
Summary of included studies reporting prevalence of one or more sleep problems in children with cerebral palsy

Author, year, country	Study design	Setting/source of recruitment	N	Gender (male)	Age (mean ± SD; range)	CP subtypes		GMFCS		Sleep measure	Comorbidities
Adiga et al., 2014,[7] India	Cross-sectional	Neurological rehabilitation department of a tertiary care hospital	50	46%	9.0 ± 2.5 y; 6.5-15 y	SD SH ST SQ DK mixed	28% 30% 8% 18% 6% 10%	I II III IV V	40% 28% 12% 2% 18%	SDSC	History of ≥1 seizure 34% (≥1 seizure in last 6 months 53%, on AEDs 65%)
Atmawidjaja et al., 2014,[36] Malaysia	Cross-sectional with control group	CP: Pediatric neurology clinic of university medical centre; HC: Nearest-age able-bodied siblings	CP: 109 HC: 109	CP: 56% HC: 51%	CP: 9.0 ± 3.9 y; 4-18 y HC: 10.0 ± 3.8 y; 4-18 y	SD SH SQ DK mixed	39% 9% 41% 9% 2%	I II III IV V	14% 17% 9% 12% 48%	SDSC	Epilepsy 52% (>4 seizures/y 53%, on AEDs 89%); severe visual impairment 22%; intellectual disability (moderate-severe) 61%
Bartlett et al., 2018,[37] Canada and USA	Cross-sectional	Multiple clinical sites across Canada and USA	671	56%	6.3 ± 2.6 y 2-12 y	SD SH SQ SO N/A	27% 28% 39% 6% <1%	I II III IV V	32% 23% 11% 18% 15%	Problems sleeping (item on the Child Health Conditions Questionnaire)	*Epilepsy 28%; visual impairment 42%; auditory impairment 13%; cognitive impairment 55%
Elsayed et al, 2013,[38] Egypt	Cross-sectional	Pediatric neurology unit of a university-based hospital	100	48%	6.1 ± 3.9 y; 2-12 y	SD SH SQ DK HYP mixed	25% 24% 15% 10% 20% 4%	I II III IV V	17% 20% 26% 10% 27%	Questionnaire on night-time sleep disorders and daytime symptoms	Epilepsy 62%; visual impairment 32%; hearing impairment 22%
Ferreira et al., 2011,[39] Brazil	Cross-sectional with control group	CP: Non-governmental rehabilitation centre; HC: age-matched sibling volunteers	CP: 90 HC: 35	CP: 64% HC: 48%	CP: 9.3 ± 3.1 y; 3-15 y HC: 8.9 ± 3.3 y; 3-15 y	SD SH SQ DK ataxic	34% 7% 34% 18% 7%	N/A		Snoring (parental report)	None had seizures ≥30 days prior to study; 36% on AEDs
Garcia et al., 2016,[40] USA	Cross-sectional	Hospital pediatric neurology, physical medicine, rehabilitation and rheumatology Clinics	73	60%	9.4 ± 4.0 y; 2-17 y	N/A		N/A		Pediatric Sleep Questionnaire	N/A
Hemmingsson et al., 2009,[41] Sweden	Cross-sectional	25 rehabilitation centres	216	N/A	mean N/A; 1-16 y	N/A (spasticity in 71%)		N/A (all used mobility aid)		Questionnaire (based on literature and sleep experts)	N/A
Horwood et al., 2018,[42] Canada	Cross-sectional	Hospital pediatric neurology clinics and national CP registry	150	61%	6.9 ± 2.9 y; 3-12 y	SD SH SQ SO DK ataxic	23% 33% 24% 7% 11% 1%	I II III IV V	44% 27% 3% 11% 15%	SDSC	Epilepsy: active 18%, controlled 15%; severe visual impairment 7%; severe auditory impairment 5%; severe cognitive impairment 14%
Koyuncu et al., 2017,[43] Turkey	Cross-sectional with a control group	CP: Children hospitalized for rehabilitation HC: N/A	CP: 94 HC: 94	CP: 55% HC: 50%	CP and HC: mean N/A; 2-17 y	SD SH ST SQ DK	33% 12% 15% 29% 12%	I II III IV V	7% 12% 40% 23% 17%	Pediatric Sleep Questionnaire	Epilepsy 20%
Maher et al., 2008,[44]	Cross-sectional	Community-based service for children with	71	65%	13.9 ± 2.0 y; 11-17 y	SD SH	44% 38%	I II	49% 27%	Self-report for 16 items on health	N/A

Australia		physical disabilities				SQ athetoid ataxic N/A	7% 3% 3% 6%	III IV V N/A	9% 13% 1% 1%	issues (2 sleep items)	
McCabe et al., 2015,[45] Australia	Cross-sectional	Home-based sleep assessment service for children with disabilities	154	54%	7.8 ± 5.4 y; 1-18 y	N/A	I II III IV V	13% 8% 11% 18% 49%	16 factors of concern related to sleep	Epilepsy 55%; visual impairment 21%; intellectual disability 44%	
Miamoto et al., 2011,[46] Brazil	Cross-sectional with control group	CP: 4 specialized institutions; HC: dental school	CP: 60 HC: 60	CP: 50% HC: 32%	CP and HC: mean N/A; 3-18 y	N/A		N/A		Sleep bruxism (proxy report and dental exam)	N/A
Ming et al., 2014,[47] USA	Cross-sectional with control group	CP: University-based pediatric neurology and pediatric epilepsy clinics and a school for children with CP; HC: university-based general pediatric clinics	CP: 52 HC: 69	CP: 56% HC: 55%	CP: 9.0 ± 7.4 y; 2-18 y HC: 9.3 ± 5.4 y 2-18 y	SD, SH, SQ, athetoid, ataxic (% N/A)		N/A		Pediatric Sleep Questionnaire	Epilepsy 56%
Mobarak et al., 2000,[48] Bangladesh	Cross-sectional	Community-based services assisting mothers of children with a disability	91	69%	3.2 ± 1.1 y; 1.5-5 y	N/A		N/A (severe gross motor disability 53%)		Behaviour Screening Questionnaire	N/A
Mol et al., 2012,[49] Belgium	Cross-sectional	4 special education schools for children with neuromotor disabilities	82	61%	9.8 ± 2.5 y; 6-15 y	SD SH SQ DK ataxic N/A	18% 24% 37% 13% 4% 4%	I II III IV V N/A	16% 28% 17% 22% 16% 1%	SDSC	Epilepsy (active) 7%; severe cortical visual impairment 15%
Munyumu et al., 2018,[50] Uganda	Cross-sectional	Hospital pediatric neurology clinic and CP rehabilitation clinic	135	49%	3.5 ± 2.0 y; 2-12 y	SH SD/SQ DK ataxic/not classified	19% 45% 19% 17%	I II III IV V	19% 19% 10% 19% 33%	SDSC	Epilepsy 33%
Newman et al., 2006,[51] Ireland	Cross-sectional	Physiotherapy department clinical database	173	58%	8.8 ± 1.9 y; 6-11.9 y	SD SH SQ DK	48% 34% 10% 8%	I II III IV V	42% 19% 17% 13% 8%	SDSC	Epilepsy 17% (33% active); severe visual impairment 6%
Radsel et al., 2017,[52] Slovenia	Cross-sectional	National CP registry	91	58%	12.3 ± 2.0 y	spastic DK ataxic	86% 13% 1%	I II III IV V	32% 22% 12% 17% 18%	Disturbed sleep (caregivers asked during registry data collection)	Epilepsy 35%; visual impairment 37% (severe 4%); auditory impairment 8% (severe 3%); IQ≤70 47%
Romeo et al., 2014a,[53] Italy	Cross-sectional	Child neurology clinics of 2 universities	165	60%	11 y (SD N/A); 6-16 y	SD SH SQ DK	23% 34% 39% 4%	I II III IV V	41% 12% 12% 10% 25%	SDSC	Epilepsy 38% (65% active, 100% on AEDs; IQ<normal/borderline 55%
Romeo et al., 2014b,[54] Italy	Cross-sectionalwith control group	CP: Neurology clinics of 2 universities; HC: Age- and gender-matched children from nurseries	CP: 100 HC: 100	CP: 52% HC: 55%	CP: 3.8 ± 0.8 y; 3-5 y HC: 3.9 ± 0.8 y; 3-5 y	SD SH SQ DK	32% 34% 29% 5%	I II III IV V	40% 23% 15% 6% 16%	SDSC	Epilepsy 32% (46% active, 100% on AED); IQ<normal/borderline 55%
Sandella et al., 2011,[5] USA	Cross-sectionalwith control group	CP: Specialized clinics, rehabilitation center websites;	CP: 41 HC: 91	CP: 56% HC: 41%	CP: 9.8 ± 1.4 y; 8-12 y	SD SH ataxic	46% 39% 7%	I II III	51% 7% 30%	Pediatric Sleep Questionnaire	History of ≥1 seizure 23%

(continued on next page)

Table 1 (*continued*)

Author, year, country	Study design	Setting/source of recruitment	N	Gender (male)	Age (mean ± SD; range)	CP subtypes		GMFCS		Sleep measure	Comorbidities
		HC: Pediatrician offices, community websites			HC: 9.7 ± 1.4 y; 8-12 y	N/A	7%	IV 7% V 5% N/A 10%			
Svedberg et al., 2008,[55] Sweden	Cross-sectional	Community-based rehabilitation centers	107	67%	11.7 ± 2.9 y; 5-13 y	spastic 77% dystonic 37%		I 37% II 19% III 4% IV 21% V 19%		Mail survey (43 items on child health)	Epilepsy 35%; visual impairment 56%; hearing impairment 5%; learning disability 38%
Zuculo et al., 2014,[6] Brazil	Cross-sectionalwith control group	CP: School clinics and health care institutions in 4 cities; HC: Age- and gender-matched, without NDDs	CP: 43 HC: 35	CP: 58% HC: N/A	CP: 9.9 ± 4.3 y; 4-18 y HC: 10.3 ± 3.7 y; 4-18 y	SD 51% SH 5% SQ 43%		N/A		SDSC, Questionnaire on Sleeping Habits & sleep diary	N/A

CP = cerebral palsy; HC = healthy controls; y = years; SD = spastic diplegia; SH = spastic hemiplegia; ST = spastic triplegia; SQ = spastic quadriplegia; DK = dyskinesia; HYP = hypotonia; N/A = not available; SDSC= Sleep Disturbance Scale for Children; AEDs = antiepileptic drugs, NDD = neurodevelopmental disorder.*Data on comorbidities and sleep outcome across GMFCS levels available for 661 children and provided by authors.

Europe (n=7),[41,49,51–55] North America (n=5)[5,37,40,42,47], and South America (n=3).[6,39,46] All studies were observational and used a cross-sectional design, with eight studies[5,6,36,39,43,46,47,54] including a healthy control comparison group. Nine[6,7,36,42,49–51,53,54] studies used the Sleep Disturbance Scale for Children (SDSC)[56] as the sleep assessment tool, four studies[5,40,43,47] used the Pediatric Sleep Questionnaire (PSQ)[57] and the remaining 10 studies[37–39,41,44–46,48,52,55] used a variety of study-specific tools to assess for sleep problems.

Sleep Assessment Tools

Sleep Disturbance Scale for Children

The SDSC is a caregiver-report questionnaire validated initially for use in children aged 6-18 years.[56] Recently in 2013, Romeo et al.[58] validated the SDSC for use in preschool children aged 3-5 years.[58] The SDSC is composed of 26 items related to the most common sleep problems found in children. The SDSC provides a total score and four subscale scores to identify disorders of initiation and maintenance of sleep (DIMS), sleep breathing disorders, disorders of excessive somnolence, and sleep hyperhidrosis. For preschool-aged children (3-5 years), the SDSC also provides subscale scores for parasomnias and non-restorative sleep; for school-aged children (6-18 years), subscale scores for sleep-wake transition disorders and disorders of arousal are calculated. Total and subscale scores on the SDSC are dichotomized as abnormal or normal using T-scores >70 or ≤70, respectively, based on published norms for children.[56,58]

Pediatric sleep questionnaire

A second caregiver-completed validated questionnaire, the PSQ, contains 22 items related to snoring, SDB, sleepiness, and behavioral problems for use in children aged 2-18 years.[57] The SDB subscale has been validated against polysomnography. Other subscores include those for daytime sleepiness and insomnia.

Other sleep assessment measures

Some of the studies[38,39,41,44–46,52,55] created their own sleep assessment tools to ascertain the presence (i.e., yes vs. no) or frequency of sleep problems. Other studies used validated general pediatric health questionnaires which included an item on sleep: the Behavior Screening Questionnaire[48] and the Child Health Conditions Questionnaire.[37] These sleep assessment tools comprised either a single question or a small number of items or involved questionnaires which were either created by the study authors or compiled from one or more existing questionnaires. In one study,[39] caregivers were questioned about children's snoring and, in another,[46] sleep bruxism was assessed using a combination of parent/sibling report and dental examinations. All sleep assessment tools relied on caregivers as informants, with the exception of one study in which adolescents self-reported on sleep quality and quantity.[44]

Quality assessment

The results of our assessment for the risk of bias are presented in Table 2.

Target population

Twenty-one of the 23 included studies[5,7,36–46,48–55] did not use representative population-based samples. Participants were recruited from a single or small number of specialized clinics or institutions,[5,7,36–40,43,46,49–51,53–55] from community-based services for children with disabilities,[44,45,48] registries,[52] or a combination of these sources.[42] Although the study by Hemmingsson et al.[41] sampled participants broadly from 25 rehabilitation centers, it was

Table 2
Assessment of risk of bias using the joanna briggs institute critical appraisal checklist for studies reporting prevalence data

Risk of bias questions	Adiga et al., 2014[7]	Atmawidjaja et al., 2014[36]	Bartlett et al., 2018[37]	Elsayed et al, 2013[38]	Ferreira et al., 2011[39]	Garcia et al., 2016[40]	Hemmingsson et al., 2009[41]	Horwood et al., 2018[42]	Koyuncu et al., 2017[43]	Maher et al., 2008[44]	McCabe et al., 2015[45]	Miamoto et al., 2011[46]	Ming et al., 2014[47]	Mobarak et al., 2000[48]	Mol et al., 2012[49]	Munyumu et al., 2018[50]	Newman et al., 2006[51]	Radsel et al., 2017[52]	Romeo et al., 2014a[53]	Romeo et al., 2014b[54]	Sandella et al., 2011[5]	Svedberg et al., 2008[55]	Zuculo et al., 2014[6]
1. Was the sample frame appropriate to address the target population?	N	N	N	N	N	N	N	N	N	N	N	N	U	N	N	N	N	N	N	N	N	N	U
2. Were sample participants sampled in an appropriate way?	N	N	N	N	N	N	Y	N	N	N	N	N	U	N	N	N	N	N	N	N	N	N	U
3. Was the sample size adequate?	N	Y	Y	N	N	N	N	Y	N	N	N	N	N	N	N	Y	N	N	N	N	N	N	N
4. Were the study subjects and the setting described in detail?	Y	Y	Y	Y	Y	N	N	Y	N	Y	Y	N	N	N	Y	Y	Y	Y	Y	Y	Y	Y	N
5. Was the data analysis conducted with sufficient coverage of the identified sample?	U	Y	N/A	U	U	N	Y	Y	N	N	N/A	U	Y	U	N	N	Y	U	U	U	U	N	U
6. Were objective standard criteria used for the identification of the condition?	Y	Y	U	U	U	Y	U	Y	Y	U	U	U	Y	U	Y	Y	Y	U	Y	Y	Y	U	Y
7. Was the condition measured in a standard reliable way for all participants?	Y	Y	Y	U	U	Y	Y	Y	U	Y	U	U	Y	U	Y	Y	Y	U	Y	Y	Y	Y	Y
8. Was there appropriate statistical analysis?	Y	Y	N/A	Y	Y	Y	Y	Y	Y	N	N	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	Y	N
9. Were all important confounding/-factors/-subgroups/-differences a) identified? b) accounted for?	Y Y	Y Y	N/A N/A	Y Y	N/A N/A	N/A N/A	Y N	Y Y	Y Y	Y N	Y Y	N N	N N	N N	Y N	Y Y	Y Y	N/A N/A	Y Y	Y Y	Y N	Y N	N N
10. Were subpopulations identified using objective criteria?	N/A	N/A	Y	U	N/A	Y	N/A	Y	Y	N/A	Y	N/A	Y	N/A	N/A	Y	N/A	Y	Y	Y	N/A	Y	N/A

N = no; Y = yes; U = unclear; N/A = not applicable.

limited to children using mobility aids. For the two remaining studies,[6,47] it was unclear if a representative sample was used as the distribution of GMFCS levels (or another validated classification tool for the degree of ambulation of children) was not provided.

Sampling

Twenty studies[5,7,36–40,42–46,48–55] were based on convenience samples which may not be fully representative of the population of children with CP. Two studies[6,47] did not fully specify how children were recruited. Only the study by Hemmingsson et al.[41], which recruited children from 25 rehabilitation centers across Sweden (questionnaire given to all eligible families) was found to have a recruitment approach that could lead to a representative sample of children with CP.

Sample size

Only three studies[36,42,50] performed a sample size calculation; each of these studies used the SDSC, and performed sample size calculations based on the study by Newman et al.[51] The smallest sample size of the three sample size calculations was reported by Atmawidjaja et al.;[36] they reported that their study required 68 children per group (CP and healthy controls) to achieve a power of 0.8 and an alpha of 0.05 based on prevalence of an abnormal total score on the SDSC of 23% by Newman et al.[51] The final sample size in the study by Atmawidjaja et al.[36] was 109 children per group. Because only these three studies included a sample size calculation, we could not evaluate the sample size in the other 20 studies. However, among the six remaining studies using the SDSC,[6,7,49,51,53,54] four studies[49,51,53,54] had samples greater than 68 children with CP (82-173 children; see Table 1). Of the 14 studies using tools other than SDSC,[5,37–41,43–48,52,55] the samples ranged from 41-661 children with CP, with eleven studies[37–41,43–45,48,52,55] having samples greater than 68 children.

Subjects and setting description

Sixteen of the studies[5,7,36–39,42,44,45,49–55] provided an adequate description of the characteristics of their samples of children with CP (see Table 1 for details).

Data analysis conducted with sufficient coverage of the sample

Four studies[41,42,47,51] carried out analyses to compare the demographic characteristics of responders and nonresponders to the questionnaires, and an additional study[36] included more than 99% of eligible children; we therefore deemed these five studies to have sufficient coverage of the sample. Three studies[44,49,55] had low response rates and did not conduct an analysis to compare the characteristics of responders and nonresponders; we deemed these studies to have insufficient sample coverage. For one study,[45] prevalence data were extracted from a retrospective review of clinical notes for all children having taken part in a home sleep assessment service, and as such we regarded this criterion as not applicable for this study. Similarly, one study[37] was part of a 5-year prospective cohort study, and thus, data from all children were included. Another study[52] had a response rate of 75%, but because children were first randomly selected from a CP registry we were not able to make any conclusions about the coverage of their sample. The remaining twelve studies[5–7,38–40,43,46,48,50,53,54] failed to provide information on response rate, nor did they describe the demographic characteristics of individuals not participating in the studies.

Criteria used for the identification of the condition

Although no studies used objective measures (such as polysomnography or actigraphy) for sleep problems, a number of studies used validated widely-used caregiver-completed questionnaires, the SDSC[6,7,36,42,49–51,53,54] and the PSQ[5,40,43,47] to assess sleep problems in children. Two studies used validated general pediatric health questionnaires the Child Health Conditions Questionnaire[37] and the Behavior Screening Questionnaire,[48] although the specific validity of these questionnaires for the assessment of sleep problems is unclear. The other studies [38,41,44,45,48,52,55] did not provide details on the validity of the questionnaires and/or specify the criteria used to ascertain sleep problems in their samples, thus uncertainty remained for this item for these studies. Similarly, Ferreira et al.[39] questioned caregivers about children's snoring and Miamoto et al.[46] assessed for sleep bruxism using a combination of parent/sibling report and dental examinations, the validity of which is unknown.

Reliable measurement of the condition

Most studies explicitly stated that the parent/primary caregiver completed the questionnaire(s),[5–7,36,37,40–43,47,49–55] or children/adolescents self-reported[44,52] in order to assess for sleep problems. For the remaining studies, the authors did not specify who provided/collected the information about sleep (the parent/primary caregiver, other family member or study staff,[38,39] or single or multiple study staff).[45,46,48]

Appropriate statistical analysis

Eighteen studies[7,36,38–43,46–55] reported prevalence values and/or provided the values needed (i.e., denominators and numerators) allowing an accurate calculation of the prevalence of sleep problems. For the study by Bartlett et al.,[37] the prevalence of sleep problems was reported for the main functional categories of children with CP only (this was the study's aim) but the authors provided us with prevalence data for the entire sample of children with CP (personal communication, Doreen Bartlett and Barbara Galuppi, 2018).

Important confounding factors/subgroups/differences (a) identified and (b) accounted for

Identified

We considered important sociodemographic and child-related characteristics that may be related to the presence, and therefore, prevalence of sleep disorders: age, CP phenotype (i.e., CP subtype and/or GMFCS level), and significant comorbidities, including epilepsy and auditory, visual or cognitive impairments. We found that 15 studies[5,7,36,38,41–45,49–51,53–55] adequately described most of these variables.

Accounted for

Among these 15 studies, only 10 accounted for more than one of these characteristics statistically, either in subgroup/subpopulation analyses for the prevalence of sleep problems,[38,45] or in logistic regression models producing odd ratios (95% confidence intervals [CIs]) for the strength of the association between factors and sleep problems,[7,36,51] or both.[42,43,50,53,54]

Subpopulations identified using objective criteria

For this item, we interpreted 'subpopulation' to mean subgroups of children with CP. We identified studies which conducted subanalyses and reported on the prevalence of sleep problems based on the sociodemographic and child-related characteristics described

previously. Twelve studies[37,38,40,42,43,45,47,50,52–55] reported on the prevalence of sleep problems in at least one subpopulation of children with CP, with five studies reporting on age,[38,42,43,45,52] two on CP subtype,[43,50] seven on GMFCS,[37,43,45,50,53–55] seven on epilepsy,[38,40,43,47,50,53,54] two on cognitive impairment,[53,54] and one on both auditory and visual impairments.[38] Among the 12 studies, only one did not describe the criteria used to define the subpopulations studied (i.e., auditory and visual impairments).[38]

Overall Quality Assessment of Studies Included in this Review

All studies included in our review included an element of selection bias (items 1 and 2 on the JBI, see Table 2). Coverage bias (item 5 on the JBI) occurred in all but five studies;[36,41,42,47,51] classification bias (item 6 on the JBI) did not occur in 13 of the studies,[5–7,36,40,42,43,47,49–51,53,54] and could not be determined in the remaining studies. Ten studies[7,36,38,42,43,45,50,51,53,54] showed a low risk of confounding bias (item 9 on the JBI). Across all 23 studies, only the studies by Atmawidjaja et al.,[36] Horwood et al.,[42] and Newman et al.[51] had a low risk of coverage, classification, and confounding bias.

Prevalence of sleep problems in children with CP

Overall prevalence of sleep problems using the SDSC

Table 3 (section A) presents the overall prevalence of sleep disorders on the SDSC for the nine studies included in the review.[6,7,36,42,49–51,53,54] Among the three studies with a healthy control group,[6,36,54] two studies[6,36] compared prevalence values between children with CP and controls (the third study compared median scores only), and only Atmawidjaja et al.[36] statistically evaluated the difference between children with CP and controls. Four studies[7,49,51,53] included school-aged children exclusively, one study[54] included preschool-aged children exclusively, whereas the remaining four studies[6,36,42,50] included children in both age groups.

Abnormal total score

Eight of the nine studies[7,36,42,49–51,53,54] reported the prevalence of an abnormal total score, which ranged from 13.0%[54] to 36.0%[7].

Age

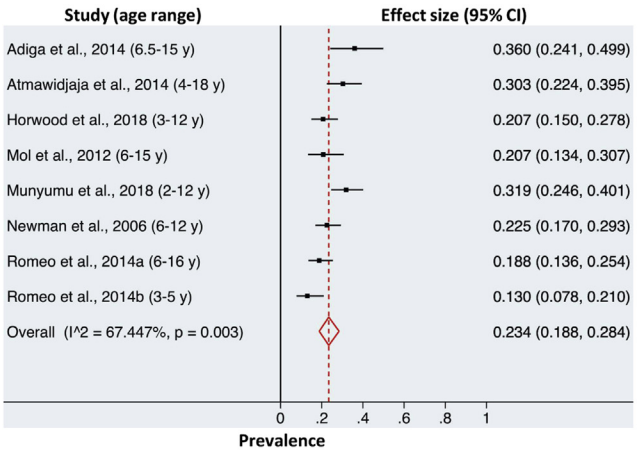
The study by Horwood et al.[42] reported abnormal total scores in 10.6% and 28.6% of preschool- and school-aged children, respectively. The two studies by Romeo et al. reported abnormal total scores in 13.0% (the lowest overall sample prevalence) and 18.8% of preschool-[54] and school-aged[53] children, respectively.

CP phenotype

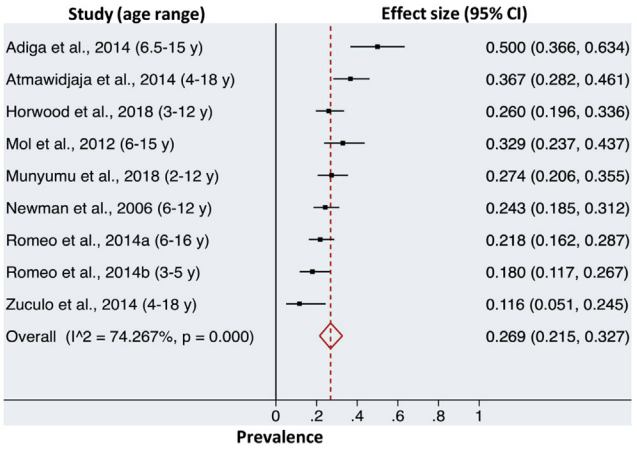
The study by Munyumu et al.[50] reported abnormal total scores in children with bilateral spastic CP (spastic diplegia/quadriplegia), unilateral spastic CP (spastic hemiplegia), dyskinesia, and ataxia/nonclassified as 41.0%, 24.0%, 30.8%, and 17.4%, respectively. This same study reported abnormal total scores by GMFCS levels I-V as 15.4%, 20.0%, 15.4%, 38.5%, and 48.9%, respectively. In their study of preschool-aged children with CP, Romeo et al.[54] reported abnormal total scores by GMFCS levels I-V as 7.5%, 4.3%, 13.3%,

Table 3
Prevalence of sleep problems in studies of children with cerebral palsy using the Sleep Disturbance Scale for Children (A) and other sleep assessment tools (B)

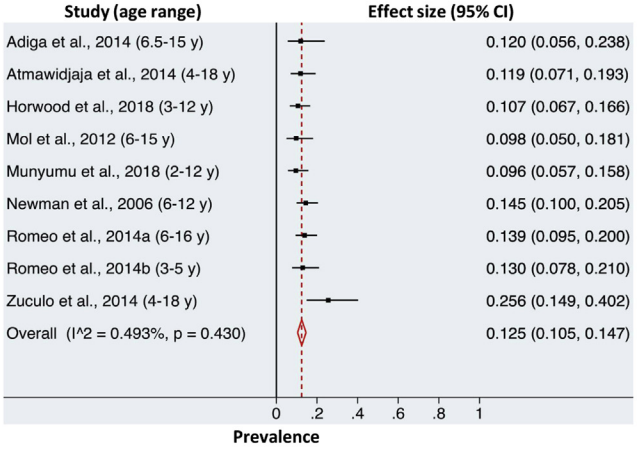
A. Sleep disturbance scale for children	
Abnormal total score	36.0,[7] 30.3,[36] 20.7,[42] 20.7%,[49] 31.9,[50] 22.5,[51] 18.8,[53] 13.0[54]
Disorders of initiation and maintenance of sleep	50.0,[7] 36.7,[36] 26.0,[42] 32.9,[49] 27.4,[50] 24.3,[51] 21.8,[53] 18.0,[54] 11.6 [6]
Sleep breathing disorders	12.0,[7] 11.9,[36] 10.7,[42] 9.8,[49] 9.6,[50] 14.5,[51] 13.9,[53] 13.0,[54] 25.6 [6]
Disorders of excessive somnolence	10.0,[7] 11.9,[36] 9.3,[42] 14.6,[49] 8.1,[50] 11.0,[51] 12.7,[53] 5.0,[54] 2.3 [6]
Sleep hyperhidrosis	6.6,[7] 5.5,[36] 8.7,[42] 2.4,[49] 10.4,[50] 5.8,[51] 7.3,[53] 9.0,[54] 34.9 [54]
Sleep-wake transition disorders	26.0,[7] 19.3,[36] 31.0,[43]11.0,[49] 13.3,[50] 17.9,[51] 15.2 [53]
Disorders of arousal	8.0,[7] 9.2,[36] 7.1,[42] 19.5,[49] 4.4,[50] 8.1,[51] 10.3 [53]
Parasomnias	6.1,[42] 9.0 [54]
Non-restorative sleep	1.5,[42] 5.0 [54]
B. Other sleep assessment tools	
a) Symptoms of disorders of initiation and maintenance of sleep	
Insomnia	36.0,[38] 89.0,[48] 78.8,[47] 41.9 [6]
Nighttime awakening	36.0,[38] 34.1,[48] 23.2 [6]
Disturbed diurnal rhythm	7.8 [41]
Insomnia, disrupted sleep and/or early wake	50.5 [55]
b) Behavioral sleep problems	
Problems in relaxing	25.0 [41]
Difficulty with settling routine	51.3 [45]
c) Parasomnias	
Sleep bruxism	38.0,[38] 23.3,[46] 27.9 [6]
Nightmares	44.0,[38] 20.8 [41]
Sleep walking	40.0,[38] 2.3 [6]
Sleep talking	8.0,[38] 11.6 [6]
Restless legs	46.0,[38] 16.3 [6]
d) Symptoms of sleep breathing disorders	
Snoring	78.9,[39] 9.6%,[43] 37.2 [6]
Sleep disordered breathing	44.0,[38] 64.4,[40] 9.7,[41] 18.1,[43] 27.9,[45] 88.5,[47] 7.3 [5]
e) Symptoms of daytime sleepiness	
Difficulty waking	18.0 [38]
(Excessive) daytime sleepiness	56.0,[38] 12.8%,[43] 36.4,[44] 63.5,[47] 48.8 [6]
f) General sleep problems unspecified	
Problems with sleeping	34.0[37]
Disrupted sleep	22.0[52]
Poor sleep quantity	11.1 [44]
Poor sleep quality	9.7 [44]



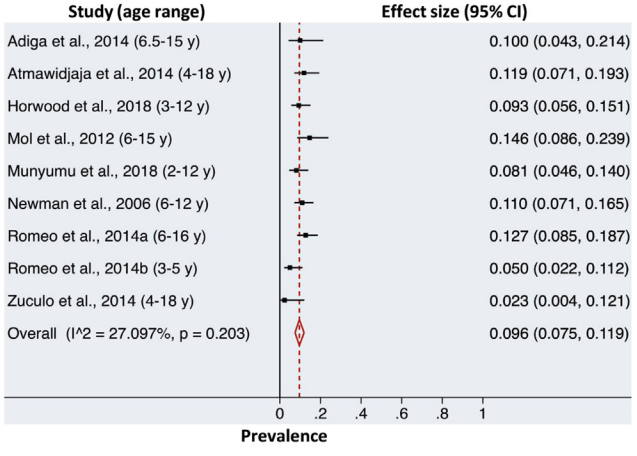
a) SDSC: Abnormal total score



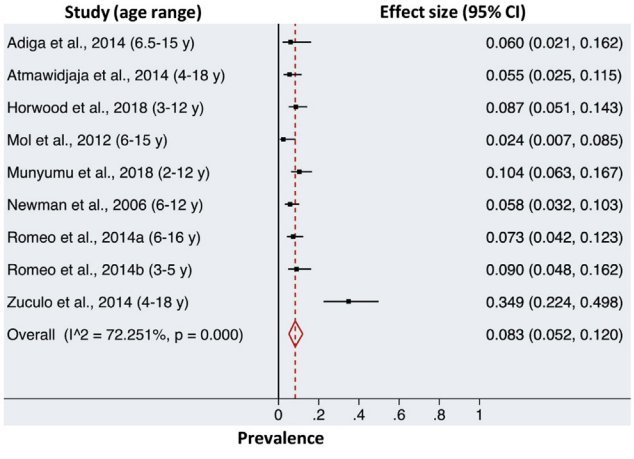
b) SDSC: Disorders of initiation and maintenance of sleep



c) SDSC: Sleep breathing disorders

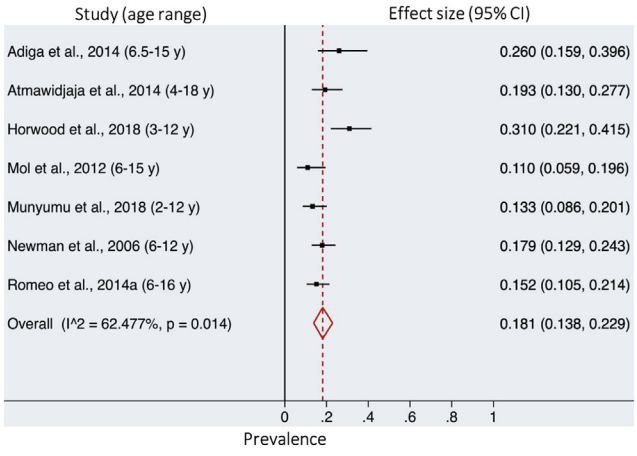


d) SDSC: Disorders of excessive somnolence

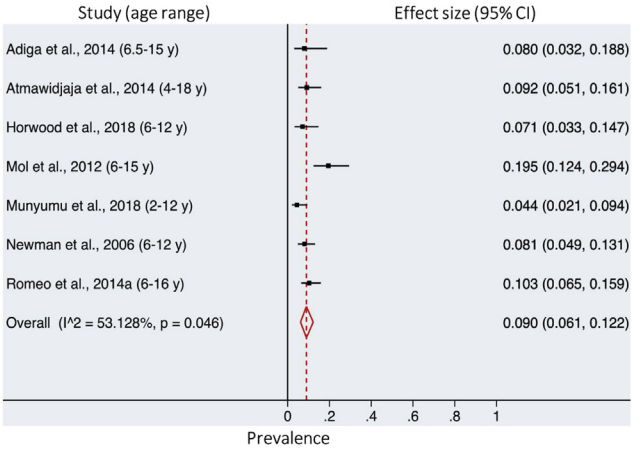


e) SDSC: Sleep hyperhidrosis

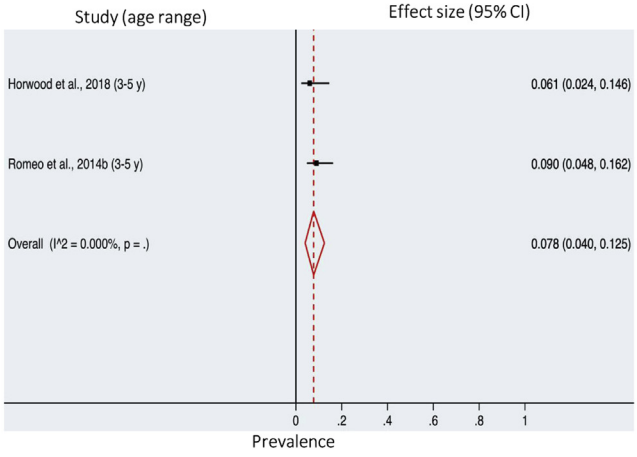
Fig. 2. Forest plots of meta-analyses for studies on the prevalence of sleep problems in children with cerebral palsy for the Sleep Disturbance Scale for Children, SDSC (a-i) and the Pediatric Sleep Questionnaire, PSQ (j). Prevalence values expressed as effect size (95% confidence interval).



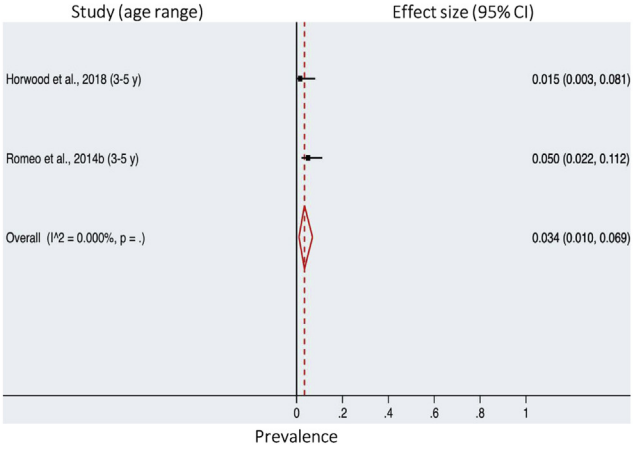
f) SDSC: Sleep-wake transition disorders



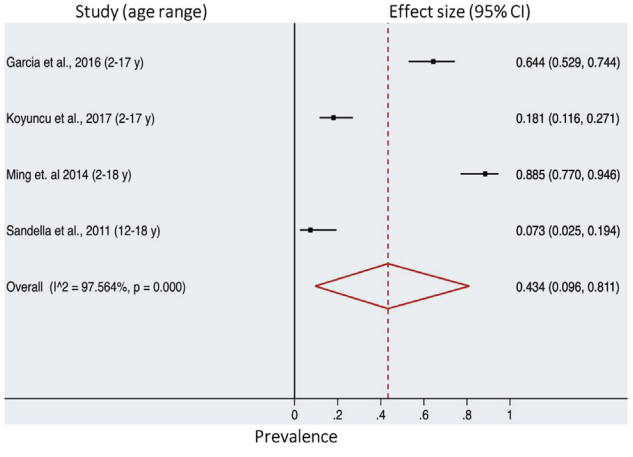
g) SDSC: Disorders of arousal



h) SDSC: Parasomnias



i) SDSC: Non-restorative sleep



j) PSQ: Sleep disordered breathing

Fig. 2. Continued.

16.7%, and 37.5%. In their study of school-aged children with CP, Romeo et al.[53] found that 12.9% of children with GMFCS levels I-IV had abnormal total scores compared with 36.6% of children with GMFCS level V.

Epilepsy

Munyumu et al.[50] found abnormal total scores in 42.2% of children with CP and epilepsy compared with 26.7% of children without epilepsy. Abnormal scores were found in preschool- and school-aged

children with active epilepsy, controlled epilepsy, and without epilepsy in 33.3%, 16.7%, and 7.5% and 34.1%, 18.2%, and 12.7% of children, respectively.

Cognitive impairment

Two studies[53,54] compared the prevalence of an abnormal total score for children with CP with (i.e., intelligence quotient [IQ]<normal/borderline) and without (i.e., IQ≥normal/borderline) cognitive impairment. The prevalence of an abnormal total score in preschool-aged children with CP and cognitive impairment was 20.0% compared with 4.4% in children without cognitive impairment.[54] In school-aged children with CP, 26.4% of children with and 9.5% of children without cognitive impairment had abnormal total scores.[53]

Comparison with controls

One case-control study reported the prevalence of abnormal total scores in both CP and control groups and found that the prevalence was significantly higher in children with CP than in controls (30.3% vs. 4.6%, p<0.001).[36]

Disorders of initiation and maintenance of sleep

All studies reported the prevalence of DIMS, which ranged from 11.6%[6] to 50.0%.[7] The study by Horwood et al.[42] reported DIMS in 18.2% and 32.1% of preschool- and school-aged children, respectively. Romeo et al.[53] found that 16.1% of school-aged children with GMFCS levels I-IV had DIMS compared with 39.0% of children with GMFCS level V. The same study reported that DIMS was found in children with active, controlled, and without epilepsy in 36.6%, 13.6%, and 17.6% of children, respectively; rates of DIMS were 27.5% in children with and 14.9% in children without cognitive impairment. Atmawidjaja et al.[36] reported that the prevalence of DIMS was significantly higher in children with CP than in controls (36.7% vs. 11.0%, p<0.001). Zuculo et al.[6] reported a lower frequency of DIMS in controls than in children with CP (5.7% vs 11.6%), but they did not test for statistical significance.

Sleep breathing disorders

All studies reported the prevalence of sleep breathing disorders, which ranged from 9.6%[50] to 25.6%.[6] The study by Horwood et al.[42] reported sleep breathing disorders in 4.5% and 15.5% of preschool- and school-aged children, respectively. Romeo et al.[53] found that 12.1% of school-aged children with GMFCS levels I-IV had sleep breathing disorders compared with 19.5% of children with GMFCS level V. The same study reported that sleep breathing disorders were found in children with active, controlled, and without epilepsy in 17.1% 18.2%, and 11.8% of children, respectively; rates of sleep breathing disorders were 3.3% in children with and 27.0% in children without cognitive impairment. Atmawidjaja et al.[36] reported that the prevalence of sleep breathing disorders was significantly higher in children with CP than in controls (11.9% vs. 5.5%, p<0.001).

Disorders of excessive somnolence

All studies reported the prevalence of disorders of excessive somnolence, which ranged from 2.3%[6] to 14.6%.[49] The study by Horwood et al.[42] reported disorders of excessive somnolence in 6.1% and 11.9% of preschool- and school-aged children, respectively. Romeo et al.[53] found that 9.7% of school-aged children with GMFCS levels I-IV had disorders of excessive somnolence compared with 22.0% of children with GMFCS level V. The same study reported that disorders of excessive somnolence were found in children with active, controlled, and without epilepsy in 24.4%, 13.6%, and 7.8% of children, respectively; rates of disorders of excessive somnolence were 15.4% in children with and 9.5% in children without cognitive impairment. Atmawidjaja et al.[36] found no significant difference between the prevalence of disorders of excessive somnolence in children with CP compared with controls (11.9% vs. 8.3%).

Sleep hyperhidrosis

All studies reported the prevalence of sleep hyperhidrosis, which ranged from 2.4%[49] to 34.9%.[6] The study by Horwood et al.[42] reported sleep hyperhidrosis in 4.5% and 11.9% of preschool- and school-aged children, respectively. Romeo et al.[53] found that 4.0% of school-aged children with GMFCS levels I-IV had sleep hyperhidrosis compared with 14.6% of children with GMFCS level V. The same study reported that sleep hyperhidrosis was found in children with absent, controlled, and active epilepsy in 9.8%, 0%, and 6.9% of children, respectively; rates of sleep hyperhidrosis were 9.9% in children with and 2.7% in children without cognitive impairment. Atmawidjaja et al.[36] found that the prevalence in children with CP was significantly higher than in controls (5.5% vs 0%, p=0.03). Zuculo et al.[6] reported much higher rates of sleep hyperhidrosis in children with CP than in controls (34.9% vs 5.7%), but they did not test for statistical significance.

Sleep-wake transition disorders

Seven of the eight studies[7,36,42,49–51,53] that included school-aged children reported the prevalence of sleep-wake transition disorders, which ranged from 11.0%[49] to 31.0%.[42] Romeo et al.[53] found that 19.4% of school-aged children with GMFCS levels I-IV had sleep-wake transition disorders compared with 26.8% of children with GMFCS level V. The same study reported that sleep-wake transition disorders were found in children with active, controlled, and without epilepsy in 22.0%, 22.7%, and 10.8% of children, respectively; rates of sleep-wake transition disorders were 23.1% in children with and 5.4% in children without cognitive impairment. Atmawidjaja et al.[36] found that the prevalence of sleep-wake transition disorders was significantly higher in children with CP than in controls (19.3% vs. 3.7%, p<0.001).

Disorders of arousal

Six of the eight studies[7,36,42,49–51,53] that included school-aged children reported the prevalence of disorders of arousal, which ranged from 4.4%[50] to 19.5%.[59] Romeo et al.[53] found that 12.1% of school-aged children with GMFCS levels I-IV had disorders of arousal compared with 2.4% of children with GMFCS level V. The same study reported that disorders of arousal were found in children with active, controlled, and without epilepsy in 4.9%, 4.5%, and 12.7% of children, respectively; rates of disorders of arousal were 7.7% in children with and 12.2% in children without cognitive impairment. Atmawidjaja et al.[36] found no significant difference between the prevalence of disorders of arousal in children with CP compared with controls (9.2% vs. 3.7%, p=0.17).

Parasomnias

In the two studies that used the SDSC in samples of preschool-aged children with CP, the prevalence of parasomnias was 6.1%[42] and 9.0%.[54]

Nonrestorative sleep

In the two studies that used the SDSC in samples of preschool-aged children with CP, the prevalence of nonrestorative sleep was 1.5%[42] and 5.0%.[54]

Sleep duration (question 1 on the SDSC)

Atmawidjaja et al.[36] reported the prevalence of various sleep durations (categories from question 1 of the SDSC) both in children with CP and controls, respectively: 9 to 11 hours (23.9% vs. 22.0%), 8 to less than 9 hours (34.9% vs. 38.5%), 7 to less than 8 hours (23.9% vs. 29.4%), 5 to less than 7 hours (10.1% vs. 9.2%), and less than 5 hours (7.3% vs 0.9%). The group found no significant difference

between the sleep duration distributions of children with CP and controls (p=0.18).

Sleep latency (question 2 on the SDSC)

Atmawidjaja et al.[36] reported the prevalence of various sleep latencies (categories from question 2 of the SDSC) both in children with CP and controls, respectively: less than 15 minutes (14.7% vs. 36.7%), 15 to less than 30 minutes (56.9% vs. 48.6%), 30 to less than 45 minutes (12.8% vs. 11.0%), 45 to less than 60 minutes (7.3% vs. 2.8%), and 60 minutes or more (8.3% vs. 0.9%). The group found a significant difference between the sleep latency distributions of children with CP and controls (p<0.001).

Meta-analysis results

Figure 2 presents the estimated pooled prevalence (random effects model) for the total score and individual sleep problems on the SDSC (forest plots a-i) and for SDB on the PSQ (plot j). Our meta-analysis for the studies using the SDSC[6,7,36,42,49–51,53,54] revealed that the most common sleep problems in children with CP were, in the descending order as follows: (1) DIMS (26.9% [95% CI 21.5-32.7%]) (2) sleep-wake transition disorders (18.1% [95% CI 13.8-22.9%]), (3) sleep breathing disorders (12.5% [95% CI 10.5-14.7%]), (4) disorders of excessive somnolence (9.6% [95% CI 7.5-11.9%]), (5) disorders of arousal (9.0% [95% CI 6.1-12.2%]), (6) sleep hyperhidrosis (8.3% [95% CI 5.2-12.0%]), (7) parasomnias (7.8% [95% CI 4.0-12.5%]), and (8) nonrestorative sleep (3.4% [95% CI 1.0-6.9%]). Among the six meta-analyses which included more than two studies, only the I^2 statistics for sleep breathing disorders and disorders of excessive somnolence had p-values >0.01. Pooled prevalence estimates for these sleep problems were considered to have insignificant heterogeneity.

Prevalence of sleep problems using other assessment tools

Table 3 (section B) categorizes the prevalence of sleep problems in children with CP as reported in studies using assessment tools other than the SDSC.

Symptoms of disorders of initiation and maintenance of sleep

Insomnia

Four studies[6,38,47,48] reported on the prevalence of insomnia in children with CP, which ranged from 36.0% in a sample of children aged 2-12 years[38] to 89.0% in a sample of children and youth aged 3-18 years.[48] Elsayed et al.[38] found that the prevalence of insomnia was significantly higher in preschool-aged children than in school-aged children with CP (46.2% vs. 25.0%, p<0.05), and reported rates of 18.2%, 31.3%, and 38.7% for subgroups of children with auditory impairment, visual impairment, and epilepsy, respectively. Ming et al.[47] reported a higher rate of insomnia in children with CP without epilepsy compared with children with CP and epilepsy and controls (82.6% vs. 75.9% vs. 46.4%). Zuculo et al.[6] found that the prevalence of insomnia was 41.9% in children with CP and 48.6% in controls (not tested statistically).

Nighttime awakening

Three studies[6,38,48] reported on the prevalence of nighttime awakening in children with CP, which ranged from 23.2%[6] to 36.0%.[38] Elsayed et al.[38] found that the prevalence of nighttime awakening was not significantly different in preschool-aged children compared with school-aged children with CP (34.6% vs. 37.5%), and reported rates of 18.2%, 31.3%, and 32.3% for subgroups of children with auditory impairment, visual impairment, and epilepsy, respectively. Zuculo et al.[6] found that the prevalence of nighttime awakening was 23.2% in children with CP and 17.1% in controls.

Delayed insomnia, disrupted sleep and/or early wake

In the study by Svedberg et al.,[55] children with CP who were identified as GMFCS IV/V compared with GMFCS I-III experienced delayed insomnia, disrupted sleep, and/or early wake a few times per month or less in 15.6% and 16.3% cases, respectively. Symptoms were experienced a few times per week or more in higher numbers of children who were GMFCS IV/V than in those who were GMFCS I-III (55.8% vs. 20.3%). Thus, these sleep problems were experienced at either frequency in 72.1% and 35.9% of children who were GMFCS IV/V and I-III, respectively.

Behavioral sleep problems

Hemmingsson et al.[41] reported that 25.0% of children and adolescents aged 1-16 years had sleep difficulties due to problems in relaxing. McCabe et al.[45] asked caregivers about their concerns regarding a variety of sleep-related factors, including difficulty with settling routines (i.e., "child does not engage in calming activities before bedtime and requires parental attention and devices as they fall asleep or settle back to sleep during the night"); we calculated an overall rate of 51.3% for this sleep problem in this study. The authors reported rates by age groups and degree of ambulation as follows: rates in children 1-5 years, 6-12 years, and 13-18 years were 68.1%, 51.3%, and 23.3%, respectively; rates by GMFCS levels (I-V) were 100%, 92.3%, 52.9%, 42.9%, and 34.2%, respectively.

Parasomnias

Sleep bruxism

Three studies reported on the prevalence of sleep bruxism in children with CP overall, which ranged from 23.3%[46] to 38.0%[38]. Elsayed et al.[38] found significantly higher rates of sleep bruxism in preschool-aged children compared with school-aged children (50.0% vs 25.0%, p<0.01), and reported rates of 18.2%, 6.3%, and 19.4% for subgroups of children with auditory impairment, visual impairment, and epilepsy, respectively. Studies including a healthy control group reported rates of bruxism in children with CP of 23.3% (vs. 15.0% in controls)[46] and 27.9% (vs. 31.4% in controls).[6]

Nightmares

Two studies reported on the prevalence of nightmares in children with CP, with rates of 20.8%[41] and 44.0%.[38] In the latter study,[38] school-aged children with CP had a significantly higher prevalence of nightmares than preschool-aged children with CP (50.0% vs. 38.6%, p<0.01), and rates of 45.5%, 56.3%, and 45.1% were for subgroups of children with auditory impairment, visual impairment, and epilepsy, respectively.

Sleep walking

Two studies reported on the prevalence of sleep walking in children with CP, with rates of 2.3%[6] and 40.0%,[38] respectively. In the study by Elsayed et al.,[38] the prevalence of sleep walking in preschool- and school-aged children with CP was not significantly different (34.6% vs. 25.0%, p=0.061), and rates of 27.3%, 18.8%, and 12.9% were reported for subgroups of children with auditory impairment, visual impairment, and epilepsy, respectively. In the latter study,[6] the reported prevalence of sleep walking in the control group was 2.9%.

Sleep talking

Two studies reported on the prevalence of sleep talking in children with CP, with rates of 8.0%[38] and 11.6%[6]. In the study by Elsayed et al.,[38] the prevalence of sleep talking was significantly higher in school-aged children than in preschool-aged children with CP

(12.5% vs. 3.4%, p<0.05), and rates of 27.3%, 18.8%, and 9.7% were reported for subgroups of children with auditory impairment, visual impairment, and epilepsy, respectively In the latter study,[6] the prevalence of sleep talking in the control group was 40.0%.

Restless legs

Two studies reported on the prevalence of restless legs in children with CP, with rates of 16.3%[6] and 46.0%.[38] In the study by Elsayed et al.,[38] the prevalence of restless legs did not differ significantly between preschool- and school-aged children with CP (42.3% vs. 50.0%), and rates of 54.5%, 12.5%, and 25.8% were reported for subgroups of children with auditory impairment, visual impairment, and epilepsy, respectively. In the latter study,[6] the prevalence of restless legs in controls was 17.1%.

Symptoms of sleep breathing disorders

Snoring

Three studies[6,39,43] reported on the prevalence of snoring in children with CP, with two studies also including rates of snoring for a control group. In the study by Ferreira et al.,[39] the prevalence of snoring in children with CP was significantly higher than that in controls (78.9% vs. 5.7%, p<0.001). In the study by Koyuncu et al.,[43] the prevalence of snoring in children with CP was no different from the prevalence found in controls (9.6% vs. 6.4%, p=0.419). Zuculo et al.[6] reported rates of snoring of 37.2% and 28.6% in children with CP and controls, respectively.

Sleep-disordered breathing (SDB)

Five studies[5,38,41,43,47] reported on the prevalence of SDB. Three of these studies[5,43,47] used the PSQ, and yielded the minimum and maximum prevalence values of 7.3%[5] and 88.5%[47] for SDB in children with CP. In the study by McCabe et al.,[45] we calculated that, overall, 27.9% of caregivers had expressed concerns regarding their child's breathing during sleep during a home-based sleep assessment; prevalence rates for this study should however be taken with caution as caregivers were asked if "the child has poorly controlled oral secretions, poor swallow, identified apneas, and history of chest infection".[45]

Age

In the study by Elsayed et al.,[38] the prevalence of SDB was significantly higher in school-aged children than in preschool-aged children with CP (50.0% vs. 38.6%, p<0.001). Koyuncu et al.[38] reported no difference in the rates of SDB between children aged 2-7 years and children aged 8-18 years (18.8% vs. 17.4%, p=0.864). In the study by Sandella et al.,[5] rates of SDB in children aged 1-5 years, 6-12 years, and 13-18 years were 25.0%, 28.2%, and 32.6%, respectively.

CP phenotype

Koyuncu et al.[43] found no difference in the frequency of SDB between children with spastic and dystonic CP subtypes (15.7% vs. 36.4%, p=0.094). The same group also did not find a significant difference between the rates of SDB by GMFCS levels (I-V: 14.3% vs. 9.1% vs. 18.4% vs. 22.7% vs. 18.8%, p=0.805). In the study by Sandella et al.,[5] reported rates of SDB by GMFCS levels (I-V) were 5.0%, 7.7%, 0%, 21.4%, and 46.1%, respectively.

Epilepsy

Garcia et al.,[40] reported that children with CP and children with CP and epilepsy had higher rates of SDB than a comparison group of children with other NDDs but neither CP nor epilepsy (58% vs. 27% and 67% vs. 27%, p<0.001 and p<0.0001, respectively). Koyuncu et al.[43] found no difference in the rates of SDB between children with CP who had or did not have epilepsy (21.1% vs. 17.3%, p=0.707). In the study by Ming et al.,[47] rates of SDB in children with CP without epilepsy, CP with epilepsy, and controls were 100%, 79.3%, and 37.7%, respectively.

Comorbidities

Elsayed et al.,[38] reported rates of 45.5%, 43.8%, and 45.1% for subgroups of children with auditory impairment, visual impairment, and epilepsy, respectively.

Comparison with controls

Koyuncu et al.[43] found that children with CP had higher rates of SDB than controls (18.1% vs. 7.4%, p=0.029). Sandella et al.[5] reported that the prevalence of SDB in controls (2.2%) was no different from that found in children with CP (7.3%).

Symptoms of daytime sleepiness

Difficulty waking

Elsayed et al.[38] reported that the prevalence of difficulty waking in preschool- and school-aged children with CP was not significantly different (11.5% vs. 25.0%), and reported rates of 9.1%, 43.8%, and 16.1% for subgroups of children with auditory impairment, visual impairment, and epilepsy, respectively.

(Excessive) daytime sleepiness

Five studies[6,38,43,44,47] reported on the prevalence of daytime sleepiness in children with CP, with a range of 12.8%[43] to 63.5%.[47] In the study by Ming et al.,[47] the prevalence of daytime sleepiness in children with CP without epilepsy, CP with epilepsy, and controls was 91.3%, 41.4%, and 27.5%, respectively. In the study by Elsayed et al.,[38] the prevalence of excessive daytime sleepiness was significantly higher in school-aged children with CP than in preschool-aged children with CP (62.5% vs. 50.0%, p<0.01), and reported rates of 9.1%, 62.5%, and 9.7% for subgroups of children with auditory impairment, visual impairment, and epilepsy, respectively. In the study by Koyuncu et al.[43] the prevalence of daytime sleepiness was not significantly different between children with CP and controls (12.8% vs. 9.6%, p=0.487). Zuculo et al.[6] reported that the prevalence of daytime sleepiness in children with CP compared with controls was 48.8% vs. 57.1%, respectively.

General sleep problems unspecified

Problems with sleeping

In the study by Bartlett et al.,[37] the prevalence of caregiver-reported problems with sleeping by GMFCS levels (I-V) was 18.9%, 30.7%, 32.0%, 40.7%, and 64.4%, respectively.

Disrupted sleep

In the study by Rasdel et al.,[52] caregivers of children (aged 8-12 years) and adolescents (aged 13-17 years) reported disrupted sleep in 18.6% and 25.0% of cases, respectively.

Poor sleep quantity

In the study by Maher et al.,[44] youth aged 11-17 years self-reported that their sleep quantity was very bad, fairly bad, fairly good ,or very good in 2.8%, 7.0%, 45.1%, and 45.1% of cases, respectively.(personal communication, Carol Maher)

Poor sleep quality

In the study by Maher et al.,[44] youth aged 11-17 years self-reported that their sleep quality was very bad, fairly bad, fairly good, or very good in 4.2%, 7.0%, 38.2%, and 50.7% of cases (personal communication, Carol Maher)

Discussion

Our systematic review aimed to synthesize the literature on the prevalence of various sleep problems in children with CP. Of the 23 studies that met our inclusion criteria, all had a cross-sectional design, and only eight [5,6,36,39,46,47,54] included a healthy control comparison group. About half of the included studies used sleep assessment tools validated for use in the general pediatric population (nine used the SDSC[6,7,36,42,49–51,53,54] and four used the PSQ).[5,40,43,47] The remaining studies[37–39,41,44–46,48,52,55] relied on a variety of unique measures with unspecified validity and reliability for the assessment of sleep problems in children. Many of the studies included a wide age range of children with CP, with most studies focusing on school-aged children and adolescents; only two studies[38,54] reported exclusively on the prevalence of sleep problems in preschool-aged children. Selection, coverage, classification, and/or confounding biases were present in all studies. Our meta-analysis showed that, among the studies using the SDSC,[6,7,36,49,51,53,54] the pooled prevalence was 23.4% (95% CI 18.8%-28.4%) for an abnormal total score, and DIMS (26.9%, 95% CI 21.5%-32.7%) was the most prevalent sleep problem reported by caregivers on this questionnaire.

Key findings from our systematic review were that older children with CP had higher rates of sleep problems than younger children with CP; children with a more severe CP phenotype had more sleep problems than children with a milder phenotype and children with CP and additional comorbidities such as epilepsy[40,43,45,50,53–55] and auditory, visual[38], and cognitive impairments[53,54] had more sleep problems than children without comorbidities. Of interest, through our review, we noted that the effects of comorbidities on the prevalence of sleep problems were not consistent. This could be in part explained by individual studies that did not adjust for all possible confounders related to sleep problems, including the comorbidities listed previously (epilepsy and auditory, visual, and cognitive impairments), in addition to socioeconomic status and genetic factors.

Seven[5,6,36,39,46,47] of the eight studies that included a healthy control comparison group provided prevalence values for one or more sleep problems, with the remaining study[54] comparing mean scores on the assessment tool between CP and controls. Only two of the studies[36,43] tested statistical differences between children with CP and typically developing children which is a shortcoming of the existing body of research on sleep problems in children with CP and limits the ability to draw evidence-based conclusions about differences between the sleep of children with CP and their typically developing peers. Atmawidjaja et al.[36] was the only group to statistically test differences in the prevalence of sleep disorders on the SDSC between children with CP and controls. The range of prevalence values for the six sleep disorders was 5.5%-36.7% in children with CP whereas it was 0-11.0% in typically developing children, with DIMS being the most common sleep problem and sleep hyperhidrosis being the least common sleep problem in both groups of children. In their comparison of rates of snoring, sleepiness, and SDB on the PSQ between children with CP and controls, Koyuncu et al.[43] found that only the prevalence of snoring was significantly different between groups (18.1% vs. 7.4%). Interestingly, Atmawidjaja et al.[36] found that, although abnormal total scores on the SDSC were six times more frequent in children with CP than in typically developing children (30.3% vs. 4.6%), the distribution of sleep duration times of the SDSC was no different between groups. In accordance with the current National Sleep Foundation sleep duration recommendations,[60] the recommended daily sleep duration varies by age group, with the least amount required among teenagers (8-10 hours) and the most among newborns (14-17 hours); yet, Atmawidjaja et al. [12] found that a large proportion (~40%) of both children with CP and typically developing children were not even getting 8 hours of sleep per day. Children with CP were found to have a longer sleep onset than typically developing children suggesting that, along with the SDSC sleep disorder prevalence values, it may be the decreased quality of sleep that differentiates children with CP from their typically developing peers.

Recently, Lélis et al.[13] published an integrative review which summarized the literature on the nature of sleep problems and associated factors in children with CP. The aims and results of our review are highly complementary as follows: our systematic review and meta-analysis provide prevalence data for well-detailed samples and subsamples of children with CP, whereas the review by Lélis et al provides information on factors reported to be associated with various sleep problems. Taken together, our reviews provide a comprehensive summary of the published evidence to date, providing important information for clinicians and caregivers regarding the types of sleep problems that may be most frequently encountered in children with CP overall. Moreover, our reviews provide information on the types of sleep problems that may be present or develop in individual children with CP based on their particular clinical profile.

The consequences of sleep disorders in children with CP can be broad and may affect both children and their families. Difficulty falling asleep and/or nighttime awakenings in children with CP may reduce and/or disrupt the sleep of their caregivers. Indeed, caregivers of children with CP who have sleep problems report experiencing a lack of sleep themselves, as well as increased stress and irritability and an extra demand on already limited personal time and energy.[7,61,62] In addition, sleep disturbances in the child with CP may lead to increased anxiety and nighttime monitoring in their caregivers, potentially reducing the quality of life of both children and their families.[63]

Sleep problems in children with CP are underrecognized and require more attention with specific questions in clinics. Once a sleep problem is identified, timely initiation of a treatment plan and access to services is crucial. As with typically developing children, the first-line treatment of sleep problems in children with CP is the promotion of developmentally appropriate healthy sleep practices and behavioral sleep interventions.[64] Behavioral sleep interventions prepare the child for sleep and promote appropriate timing, duration, and effectiveness of sleep.[65] Pharmacological treatments may be an option for sleep problems not amenable to behavioral interventions. The timely identification and treatment of sleep problems may improve other related health outcomes, such as physical development and growth, neurobehavioral functioning, and the quality of life of both children with CP and their families. Though treatment interventions were beyond the scope of our systematic review, this would be a subject of interest for future systematic reviews.

As we report in our quality assessment, the studies included in our systematic review had some notable limitations. No studies used polysomnography, the gold standard for the assessment of sleep disorders (in particular sleep breathing disorders), and no studies used actigraphy, an objective device that quantifies periods of sleep and wakefulness. Although actigraphy in children with CP is more challenging than in typically developing children, actigraphy has been shown to be feasible and acceptable in children with CP. In children with unilateral limb involvement, the actigraph can be placed on the less paretic arm. Underhill et al showed that wrist actigraphy was successful in children with CP of all GMFCS levels.[66] In children who have bilateral limb involvement or children who are not able to tolerate the wrist placement, Adkins et al have shown that an alternate method would be to place the actigraph on the shoulder.[67]

All studies included in this review assessed children's sleep problems based on proxy/caregiver reports, which may overestimate or underestimate the true prevalence of sleep problems in children with CP. More than half of the studies did however use the SDSC and PSQ, which are validated questionnaires for use in typically developing children, but not children with CP specifically.[68] Most

studies recruited children through clinics and other specialized services for children with NDDs and thus included largely referred samples that may not represent the broader population of children with CP, particularly those children who are high-functioning and do not require specialized medical services. Future studies utilizing population-based registries of children with CP will provide more accurate prevalence data on sleep problems and other clinical parameters. Some studies provided limited or no information on important clinical descriptors such as CP phenotype and important comorbidities that, as highlighted in the review by Lelis et al.[13] and studies included in our systematic review, could be associated with higher rates of specific sleep problems. Taken together, these shortcomings in the quality of the individual studies reporting on the prevalence of sleep problems in children with CP may limit the generalizability of their findings to the overall population of children with CP.

Our systematic review also has a few limitations. There may be publications that were not identified in our review, although we attempted to search and include all relevant literature through an extensive search strategy developed in collaboration with an experienced medical sciences librarian, searching through several electronic databases, and review of publications in two languages (English and French). Most studies included in our systematic review drew conclusions from relatively small, clinic-based and thus potentially biased samples; the interpretation of our results should therefore be taken with caution. For our meta-analysis, we were limited in two cases by the number of retrieved studies (i.e., our estimate of the pooled prevalence of parasomnias and nonrestorative sleep was based on only two studies), again suggesting that future well-designed studies on sleep problems in children with CP are needed to develop a broader literature base from which to derive clinical conclusions.

Conclusions

The prevalence of sleep problems in children with CP is high (23.4% [95% CI 18.8-28.4%]) based on our meta-analysis of eight studies reporting an abnormal total score on the SDSC. Although within a similar range as typically developing children, the prevalence is lower than the prevalence of sleep problems in children with other NDDs (autism spectrum disorder: 50-80%;[69] attention deficit hyperactivity disorder: 35-70%).[70] However, there is notable variability in the prevalence of sleep problems between subgroups of children with CP. Children with a severe CP phenotype (e.g., nonambulatory/GMFCS level IV-V and/or quadriplegia) and/or those with certain associated comorbidities (e.g., epilepsy) are at increased risk of sleep problems compared with those with a milder phenotype (e.g., ambulatory/GMFCS levels I-III and/or hemiplegia) and those without comorbidities. The limited evidence in our review (two studies using validated sleep assessment tools and a healthy control group) suggests that the prevalence of sleep problems in children with CP is significantly higher than the prevalence of sleep problems in their typically developing peers. Future studies in large groups of children with CP using validated measures, including objective tools, and which include control groups of typically developing children and more detailed subgroup analyses are recommended. Understanding the extent of sleep problems and associated risk factors will help design interventions to improve the sleep of children with CP.

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