



## List of Abbreviations

CP	Cerebral palsy
CSHQ	Children's Sleep Habits Questionnaire
DIMS	Disorders of initiation and maintenance of sleep
EDS	Excessive daytime sleepiness
EEG	Electroencephalogram
GMFCS	Gross Motor Function Classification System
NDDs	Neurodevelopmental disabilities
OSA	Obstructive sleep apnea
PSG	Polysomnography
PSQ	Pediatric Sleep Questionnaire
QOL	Quality of life
REM	Rapid eye movement
SDB	Sleep-disordered breathing
SDSC	Sleep Disturbance Scale for Children
SWTD	Sleep-wake transition disorders
TD	Typically developing

## Introduction

Cerebral palsy (CP) represents the most frequent cause of physical disability in childhood, affecting more than 2 per 1000 live-born children [1]. It consists of a group of disorders of movement and posture caused by a nonprogressive interference, lesion, or abnormality of the immature brain, which is often accompanied by impairment of sensation, cognition, and communication [2]. In recent years, interest has grown in the description and diagnosis of sleep disorders in children with CP, with reports that sleep disorders are more frequent in CP than in typically developing (TD) children [3–19]. The prevalence of sleep disturbances in CP varies from 19% to 63% according to different studies, which

have involved different types of sleep disorders and ages of subjects and seem related to a multifactorial etiology including muscle spasms, musculoskeletal pain, decreased ability to change body position during the night, visual impairment, epilepsy, and side effects of antiepileptic drugs [3–7]. Other comorbidities, such as motor and cognitive impairment and psychiatric problems, increase the risk of developing abnormal patterns of sleep [6–9]. As the presence of sleep disorders influence the quality of life of all the family [10, 11], correct diagnosis and treatment could improve not only the well-being of children with CP but also of their caregivers. The focus of the present chapter is to provide a critical overview of sleep disorders associated with CP with a specific focus on the diagnosis and treatment.

## Case Report

Antonio is a 6-year-old boy with cerebral palsy and sleep disturbance. Born at 31 weeks' gestational age, he developed periventricular leukomalacia and spastic diplegia with motor and cognitive impairment and behavioral issues. He is classified at a Level 1 on the GMFCS and has a full-scale IQ of 60. On the Child Behavior Checklist, he showed mainly externalizing disorders (aggressive behavior and attention problems). No epilepsy or clinical seizures have been reported. His parents reported significant sleep issues, mainly difficulties in getting to sleep at night, feeling afraid when falling asleep, and waking up several times per night. He completed the Sleep Disorders Scale for Children (SDSC), a questionnaire specifically designed to assess sleep disturbances in children, with the following results: Total Score 85, Disorders of Initiating and Maintaining Sleep 76, Sleep Breathing Disorders 79, Disorders of Arousal 70, Sleep-Wake Transition Disorders 70, Disorders of Excessive Somnolence 77, and Sleep Hyperhidrosis 57. He had a normal sleep electroencephalogram (EEG).

Parents were advised to promote a good bedtime routine and to provide a dark and quiet environment, reducing

E. Mercuri (✉) · D. M. Romeo  
Pediatric Neurology Unit, Policlinico Gemelli, Catholic University,  
Rome, Italy

activities such as watching television during the lead-in to bedtime. After 2 months of specific behavioral intervention with parent training, the parents reported an improvement in sleep behavior, but the child continued to wake up at least two to three times per night; melatonin was started with a dosage of 2.5 mg, 20–30 min before bedtime. Parents reported a further improvement of quality and quantity of sleep, especially in sleep latency and nighttime sleep duration, with a notable reduction in the nocturnal episodes of waking up (down to one to two times per week). After 3 months the parents completed again the SDSC with the following results: Total Score 58, Disorders of Initiating and Maintaining Sleep 60, Sleep Breathing Disorders 58, Disorders of Arousal 58, Sleep-Wake Transition Disorders 45, Disorders of Excessive Somnolence 53, and Sleep Hyperhidrosis 57. No side effects of melatonin were reported.

## Prevalence

There is no consensus on the prevalence of sleep disorders in children with CP, due to heterogeneity of the types of sleep disorder assessed, methods of assessment, and the presence of other comorbidities. Published data using screening questionnaires reflecting the overall quality of sleep in children with CP [5–7, 12, 19, 20] identified an abnormal total sleep score in 19–30%, with at least one clinically significant sleep disorder in 40–63% at school age, and a lower prevalence in preschool children. In terms of specific sleep disorders, disorders of initiation and maintenance of sleep (DIMS), sleep-wake transition, excessive daytime somnolence (EDS), and arousal, as well as sleep-disordered breathing (SDB), were all reported with a prevalence between 10% and 24%. Difficulties in settling a child to sleep might lead to actions that play a role in perpetuating insomnia, such as rocking or patting [20]. In another study [19], the most frequent sleep disorders reported were bruxism (32.8%), leg movements (29.5%), nocturnal enuresis (24.6%), and sleep-disordered breathing symptoms like snoring (36.1%). Furthermore, obstructive sleep apnea (OSA) and habitual snoring have also been reported in children with CP, potentially reaching a very high incidence (>60%) [21]. DIMS is more frequent in children with spastic quadriplegia, dyskinetic CP, or severe visual impairment [5]. In terms of severity of motor function as measured by the Gross Motor Function Classification System (GMFCS), insomnia and sleep-disordered breathing (SDB) were more common in children with grades V and IV (severe motor impairment), bruxism was more common in GMFSC grade III, and nightmares and sleepwalking were more common in GMFSC grades I and II (mild motor impairment) [1].

## Etiology

Sleep disorders in children with CP can be related to the primary motor impairment due to muscle spasms, musculoskeletal pain, and decreased ability to change body position during the night, but other risk factors can be identified.

*Epilepsy* is well known to predispose to sleep disorders [22, 23]. It is found in approximately 50% of children with CP. The interaction between epilepsy and sleep disorders is not completely understood. Different studies have confirmed that sleep problems were more prevalent in children with active epilepsy; conversely, sleep disturbances, especially sleep deprivation predispose to an increase in the frequency of seizures [22, 23]. These effects vary considerably with the type of seizure disorder, including its etiology and comorbidities. Direct effects are most likely in severe forms of epilepsy with frequent, difficult to control seizures especially of a convulsive type [22]. Nocturnal seizure discharge in temporal/frontal lobe epilepsy can cause parasomnias interfering with nighttime sleep structure and causing excessive daytime somnolence and worsening sleep apnea [21–23].

Sleep organization in children with CP was characterized by abnormal sleep EEG, sometimes with the absence of rapid eye movement (REM) sleep, low incidence of sleep spindles, or high percentage of wake after sleep onset [16]. Antiepileptic drugs can affect sleep quality and daytime alertness and could modify sleep architecture. However, some studies demonstrated that sleep disorders could be more associated with persistent seizures than with antiepileptic drugs [3–7]. In these studies, seizure-free epileptic children, who were all receiving antiepileptic treatment, reported no increase of excessive daytime somnolence. On the other hand, daytime drowsiness was much more strongly associated with persistent seizures than with antiepileptic drugs, perhaps due to a disruptive effect of seizures on sleep physiology or to an expression of postictal effects.

*The severity and type of motor impairment* is also strongly associated with specific sleep disorders. Children with spastic quadriplegia and with dyskinesia were more affected by DIMS, due to pain related to stiffness and contractures or involuntary movements. Motor problems of the dyskinetic form of CP could lead to motor restlessness during sleep, linked to a dopaminergic dysfunction or to basal ganglia lesions that could account for hyperkinesia, hypnic jerks, and bruxism [24, 25]. On the other hand, the brainstem dysfunction described in children with dyskinetic CP may affect the architecture of sleep, potentially in disturbances of REM sleep [8].

*Brain lesions, especially if affecting cortical and subcortical structures related to the central visual pathway, can cause impairment of several aspects of visual function, such*

as *visual fields or acuity*. This is described as *cortical visual impairment* and occurs in approximately 20–50% of children with CP. It has been reported that concomitant visual impairment in children with CP can also affect the timing and maintenance of sleep through the lack of light perception resulting in altered melatonin secretion and potentially a free running circadian rhythm [4]. Furthermore, the presence of behavioral problems in these children could contribute to sleep wake cycle disorders, as reported in a recent study showing a positive correlation between difficult morning awakening and visual impairment [19].

*Behavioral and psychological problems* are also known to affect the quality of sleep both in children and their families. Several studies reported a high incidence of behavioral problems in children with CP, such as aggression and attention problems or withdrawal and somatic complaints, and the association between behavior and sleep disorders has largely been reported even in children without disabilities [26–28]. Two studies [6, 7] reported a specific association between sleep disorders and behavior in children with CP both in preschool and elementary school, confirming that behavioral problems were often associated with abnormal sleep disorders, especially internalizing disorders such as withdrawal and somatic complaints. In a large study of preschool children with CP, specific sleep disorders such as SDB, bruxism, and EDS were associated with psychiatric problems [19]. Another study related sleep problems in children with CP to maternal depression, showing that mothers who had children with sleep problems were more likely to have difficulty sleeping themselves [11]. Insomnia, excessive daytime sleepiness, and GMFCS score severity were associated with lower quality of life (QOL) in children with CP [10]. Sleep disturbances in children with CP influence not only their caregivers' sleep but also their siblings', who show shorter sleep and more difficulty falling asleep compared to their peers [29]. In a study exploring sleep disturbances in children with CP and their siblings using a structured questionnaire, children with CP scored significantly higher than their siblings in most of the scores, but siblings reported a higher prevalence of DIMS than children in the general population. Furthermore, a high/abnormal total score in CP was significantly associated with reduced sleep duration and increased sleep latency in caregivers [30].

*Anatomical factors*, as glossoptosis, adenotonsillar hypertrophy, recurrent aspiration pneumonia, and gastroesophageal reflux, are reported in children with severe CP and could contribute to sleep-related breathing disorders, causing sleep fragmentation and hypoxemia [3, 5]; habitual snoring and sleep apnea have been reported with a high prevalence in children with CP [5, 21, 31].

## Diagnosis

Several methods have been proposed to assess sleep disturbances in children with CP to investigate developmental changes in sleep behaviors and to identify differences between TD children and those with CP.

### Polysomnography (PSG)

Polysomnography (PSG) is considered the gold standard sleep measurement [32]. According to international guidelines [33], electrodes are placed at specific locations on a child's body (scalp, face, neck, chest, and legs), for continuous electrophysiological recordings of brain activation, eye movements, skeletal muscle activation, and cardiac function. In addition, respiratory monitoring is measured using an oronasal thermal sensor for airflow, esophageal manometry or more commonly, respiratory inductance plethysmography for respiratory effort, pulse oximetry for oxygen saturation, and transcutaneous or end-tidal PCO<sub>2</sub> monitoring to measure hypoventilation. These electrophysiological data provide a range of information about sleep: how long the child takes to fall asleep, total sleep time, how well the child slept overall, nocturnal awakenings, limb movements during sleep, and breathing difficulties [32]. PSG has been used in few studies of children with CP especially to assess OSA and to follow improvement after specific treatments [34–36]. In children with severe CP [9], obstructive apnea, decreased ability to change body position, and interictal epileptiform discharges are prevalent during sleep, with significantly more respiratory disturbances per hour of sleep, fewer changes in body position during the night, and interictal epileptiform discharges averaging 23.3% of total arousals.

The limitation of this method in routine clinical practice is that it requires placement of electrodes by a specialist and usually requires that a child stay overnight in a sleep laboratory, a procedure which is costly and inconvenient for some families.

### Actigraphy

Actigraphy is an increasingly popular method of assessing habitual sleep-wake patterns, using a watch-like movement sensor to assess activity levels as a proxy for likely sleep and wake [32]. It allows data collection over different days for sleep measurement within a child's natural environment; therefore, compared to PSG, actigraphy is less costly and invasive than PSG. Actigraphy data provide information about the length of a child's sleep, whether he experienced

any awakenings, and how efficient his sleep was overall. Actigraphy is not a suitable method for the diagnosis of disorders in which sleep is fragmented: for example, the detection of limb movement events in children with periodic limb movement disorder is not accurate. Furthermore, in children with OSA, actigraphy fails to reliably identify breathing abnormalities, while PSG is the gold standard for diagnosis [32]. Recently it has also been validated to monitor physical activity during walking in children with CP and could be used in rehabilitation research and clinical practice [37].

### Sleep Electroencephalography (EEG)

EEG has been used to measure sleep activity both in clinical and research settings in children with neurodevelopmental disabilities (NDDs) [32]. Specific age norms in sleep EEG could be useful in identifying NDDs: slow-wave activity during sleep increases over the first years of life with a peak before puberty, followed by a decline during adolescence, as it is generated and maintained by thalamocortical and cortico-cortical networks. Children with CP who have lesions affecting these networks could be identified by the assessment of sleep EEG. The same findings could be identified for sleep spindles, a characteristic feature of non-rapid eye movement (NREM) sleep that is related to the same network activities. In one study, more than 50% of children with CP and mental retardation showed the absence of NREM sleep and REM sleep, either extremely low incidence of sleep spindles or the presence of extreme spindles or an abnormally high percentage of wake after sleep onset [16]. Using a compressed spectral array during nocturnal sleep, researchers also observed periodic changes of delta (slow wave) and spindle rhythm powers related to intelligence quotient, with a significant decrease in the developmental quotient in CP patients with neither delta nor spindle rhythm powers found during nocturnal sleep [38]. Furthermore, among people with athetoid CP [8], there has been reported a marked decrease in rapid eye movements during REM sleep.

### Questionnaires and Sleep Diaries

Both types of instruments are widely used to ask parents or children to reflect on daily or weekly sleep behavior. Questionnaires are easy to administer and straightforward to score and sometimes report normal values or ranges for comparative purposes. Diaries require a daily report of sleep and wake times [32]; they are time-consuming to complete, and their results can be difficult to interpret, as there are no normal values or times that exist for comparison. Clinicians frequently opt to use questionnaires instead of PSG and/or

actigraphy because of their time- and cost-effective nature, as well as their relative ease of administration, but these may sacrifice accuracy of diagnosis for both SDB and DIMS. Several standardized scales are available to identify sleep disorders in childhood, but only few have been used in children with CP [39–41].

The Pediatric Sleep Questionnaire (PSQ) [39] is mainly used to screen for childhood sleep-related breathing disorders; it contains 48 items related to OSA symptoms. Subscales include the Sleep-Related Breathing Disorder scale, a measure of SDB. Furthermore, it has been used to study the association between quality of sleep and QOL in 41 children with CP and in 91 TD controls age 8–12 years [10], reporting risks for sleep disruption and lower QOL in children with CP; insomnia was often associated with a low psychosocial QOL, whereas excessive daytime sleepiness predicted lower physical QOL. On the other hand, no effects of sleep variables on QOL were observed among the TD group.

The Children's Sleep Habits Questionnaire (CSHQ) is designed for school-age children, and it is one of the most common tools used for assessing sleep problems in children [40]. It is an up to 45-item, parent-rated questionnaire that assesses the frequency of behaviors associated with common pediatric sleep difficulties as they have occurred during a "typical" recent week. Items cluster into eight subscales that relate to common sleep problems in children: Bedtime Resistance, Sleep-Onset Delay, Sleep Duration, Sleep Anxiety, Night Wakings, Parasomnias, Sleep-Disordered Breathing, and Daytime Sleepiness. In addition, all ratings are summed to create a total sleep disturbance score, for which a score of over 41 identified 80% of a clinical sleep disorders sample and is often used as a cutoff for abnormality. A study comparing CSHQ scores between 40 children with CP, aged 4–12 years, and 102 age-matched TD children showed that children with CP had higher scores for Sleep Anxiety, Night Wakings, Parasomnias, and SDB subscales [11].

The Sleep Disturbance Scale for Children (SDSC) [41] looks at sleep disorder symptoms over the previous 6 months in children between 3 and 16 years [42]; it consists of 26 items in a Likert-type scale with values 1–5 (higher numeric values reflect higher severity of symptoms). The sum of scores provides a total sleep score with a possible range from 26 to 130; a T-score of more than 70 (>95th centile) is regarded as abnormal and a score <70 or less as normal. The original factor analysis yielded six sleep disturbance factors representing the most common areas of sleep disorders in childhood and adolescence: (a) DIMS, (b) sleep breathing disorders (SBD), (c) disorders of arousal (sleepwalking, sleep terrors, nightmares), (d) sleep-wake transition disorders (SWTD) (hypnic jerks, rhythmic movement disorders, hypnagogic hallucinations, nocturnal hyperkinesias, bruxism),

(e) disorders of excessive somnolence, and (f) sleep hyperhidrosis. The SDSC represents the most frequently used questionnaire in studies of sleep in children with CP, with more than 700 children [5, 7, 11, 13, 20] assessed in different studies. Compared with normative data, children with CP showed higher total SDSC scores, with specific sleep disorders as DIMS, SDB, and SWTD; the sleep disorders were closely correlated to motor and cognitive impairment, behavioral problems, and epilepsy.

## Treatment

Recent reviews have confirmed the absence of studies on sleep intervention specifically for children with CP [3, 15]. Most of the published research on intervention has been related to other NDDs, with only a few including participants with CP [15]. In these studies, it is recommended that the treatment for sleep disorders should begin by establishing good “sleep hygiene,” through parent-based education and behavioral interventions [3, 43, 44]. This should be followed by promoting a structured, age-appropriate bedtime routine and providing a dark and quiet environment. Potentially stimulating activities such as watching television and vigorous play should be avoided during the lead-in to bedtime. Maintaining regular bedtime and waking times may strengthen and entrain circadian mechanisms to promote rapid sleep onset near the desired bedtime.

Behavioral interventions adopted by parents should be used in case of disruptive behaviors [43, 44]: *extinction (systematic ignoring)*, placing children in bed and then ignoring (directly or gradually) any inappropriate behaviors, with the exception of concerns regarding safety or illness; *positive routines*, creating quiet but enjoyable bedtime routines with fading of bedtimes toward children’s time of habitual sleep onset; and *parent education and anticipatory guidance*, educating parents and caregivers about early childhood sleep and the treatable influences that may promote or disrupt it.

## Osteopathy and Massage

These therapies are among of the most used alternative medicine therapies for children with CP. The effects of osteopathy on sleep disorders in CP have been evaluated in two studies [45, 46]. In the first, the authors assessed 142 children with CP, 71 of whom were treated with osteopathy, for 6 months. No statistical differences were found for motor function, quality of life, pain, or sleep between children who had cranial osteopathic treatment and those who did not [45]. In the second study, the authors treated 50 children with CP using either osteopathic manipulation or acupuncture [46]; after a period of 6 months, 96% of the parents reported an

improvement, especially in the use of arms or legs, and a more restful sleep in both the osteopathic and acupuncture groups. In both studies, however, no structured questionnaires or other sleep measures as actigraphy or PSG were used.

The literature on the use of massage specifically for children with CP is sparse and controversial: some studies supported the benefit of massage therapy for spasticity and also improved muscle tone, range of motion, and cognition [47], while others failed to find any benefit [48]. In a recent study, 100 families of children with CP responded to a survey documenting that 80% of their children with CP had received massage at least once. Of these families, 86% reported that massage helped to relax their children’s muscles, 71% that it improved quality of life, 30% that it decreased their child’s pain, and 23% that it improved sleep [49]. An improvement in sleep behavior was reported in another study enlisting 70 parents in a parent-training program for massage in children with CP, with improvements in eating and mobility also claimed [50]. However, neither study used a control group, which limits more definitive conclusions.

## Surgery Treatment

Children with CP are commonly affected by OSA, due to anatomical factors that lead to airway obstruction [3–5, 17, 21]. Different surgical techniques, such as adenotonsillectomy, soft-tissue reduction combined to skeletal expansion of the mandible, uvulopalatopharyngoplasty, and tongue-base suspension, have led to significant improvement of OSA in children with CP, with reduced apnea/hypopnea indices and improved oxygen saturation [17, 35, 51, 52]. However, in the case of severe airway obstruction, especially at older ages, children with CP could require tracheostomy [53].

## Treatment for Spasticity

Baclofen and botulinum toxin are medications frequently used for spasticity in children with CP. Over the last few years, some studies have also explored a possible role of these treatments in improving sleep behavior. Intrathecal baclofen delivered by implanted pumps has been demonstrated to reduce the frequency of night wakings and sleep apnea and to improve sleep in general in children with spastic CP within 6–9 months after treatment, probably due to the resulting reduced muscle spasms and improvement of pain and mobility [18, 54, 55]. Rare and generally temporary side effects, such as constipation, excess lethargy, urine retention, pressure sore, and deteriorating swallow, were reported.

The effect of botulinum toxin injections on sleep has been studied in 26 children with non-ambulatory quadriplegic CP

who had significant spasticity and pain at the hip level [56]. These subjects received botulinum toxin injections at the adductor magnus, medial hamstrings, and iliopsoas muscles. All the children reported significant improvement at the Pediatric Pain Profile after 3 months of the treatment; furthermore, families reported an improved sleep pattern of the child, especially in terms of the reduction of the frequency of night wakings and need for turning because of sleep discomfort. No significant side effect was reported after treatment.

## Melatonin and Other Drugs

Drug treatment of sleep disorders in children has received remarkably little scientific study, and there is a lack of well-designed controlled studies, despite widespread use in this age group [43]. Melatonin (N-acetyl-5-methoxytryptamine), a tryptophan-derived molecule, is a chronobiotic drug essential for the regulation of the sleep-wake cycle. During the day, human synthesis of melatonin is limited. When it becomes dark, norepinephrine liberated by the beta-adrenergic receptors in pinealocytes improves its secretion [3, 43]. It has been proposed that melatonin may have significant therapeutic effects in circadian rhythm sleep-wake disorders such as jet lag disorder and sleep-wake cycle disturbances in blind people and shift workers. There are various studies of melatonin usage in pediatric sleep disturbances, most of them confirming its role in improving sleep quality, reducing sleep-onset latency, and increasing total sleep time [3, 57–63], but none has included exclusively CP patients. Some studies have reported results on the use of melatonin in NDDs, including CP [58–63]. Wasdell et al. [58] used a starting 5 mg dose of melatonin in 51 children affected by different NDDs, with a gradually increased increment of the dose until the therapy showed optimal beneficial effects. Sleep characteristics were measured by wrist actigraphs and caregiver completed diaries. The children showed an improvement in both total nighttime sleep and sleep latency of approximately 30 min. Significant improvement in children's sleep was also observed in clinician and parent ratings.

In a randomized, double-blind, placebo-controlled 6-week trial of melatonin versus placebo in 20 children with NDDs, 90% fell asleep more quickly when receiving melatonin than placebo with a significant reduction of sleep latency [59]. In a third study, melatonin was used in 49 children aged 1–13 years affected by CP, epilepsy, and other NDDs; children under 5 years of age were started on 2.5 mg, and those  $\geq 5$  years started on 5 mg; this was increased by 2.5 mg at intervals of 3 days up to a maximum dose of 7.5 mg (<2 years) or 10 mg. Parents completed sleep diaries before and after treatment, and the majority showed significant improvement in total sleep time and in nighttime sleep duration and sleep

latency [60]. A recent clinical trial confirmed the efficacy of melatonin in the treatment of sleep disorders in 275 children with NDDs, with doses ranging from 0.5 to 12 mg for a period of 12 weeks, again reducing sleep latency and increasing total sleep time [61].

As melatonin is a generally safe treatment with few adverse side effects reported, it could therefore be considered a first line of treatment of sleep disorders in children with CP, especially involving sleep onset [63]. As it has been reported that in a limited number of cases of severe neurologically disabled children melatonin can potentially increase seizure activity [62], there has been some discussion on whether its use should be limited in children with CP with comorbid active epilepsy [15]. Across disorders, additional research including larger groups of children with specific disabilities/syndromes is needed to draw disability-specific conclusions [63].

Gabapentin is an antiepileptic medication whose mechanism of action has not fully been described but may involve interactions with voltage-gated calcium channels and modulation of GABA biosynthesis. It has been used for the treatment of partial seizures, neuropathic pain, and restless legs syndrome. In adults with primary insomnia, gabapentin improves sleep quality, increasing slow-wave sleep and sleep efficiency [64]. Recently it has been used in treating refractory insomnia in 23 children with a mean age of 7 years and a diagnosis of NDDs in 87% (including one child with CP); gabapentin was started at an average dose of 5 mg/kg, with a maximal dose of 15 mg/kg at bedtime; at follow-up the 78% of the children showed a sleep improvement, with adverse effects in 6 children [65].

---

## Conclusions

Sleep disorders are common in children with cerebral palsy. Its characteristic and comorbid features, such as motor impairment, pain, cognitive impairment, behavioral problems, or epilepsy, are all important risk factors for the development of sleep disorders. The use of questionnaires, primarily the SDSC to date, is widely reported to identify sleep disturbance, even if a more structured in-depth interview or objective assessment, using PSG or actigraphy, provides better information on sleep disorders and allows a more accurate diagnosis.

No sleep interventions specifically designed to improve the sleep of children with CP are reported in the literature. Instead, strategies to improve sleep depend mainly on clinical experience and include sleep hygiene and/or specific drugs such as melatonin, a commonly prescribed drug for disturbed sleep in children with neurological dysfunction. An effective treatment has the potential to improve not only the well-being of the child but the well-being of a whole family [3–5, 15, 29, 30].

## Future Directions

Randomized placebo-controlled trials should be proposed to establish efficacy, safety, and pharmacokinetics data of melatonin in children with CP to test the hypothesis that oral melatonin has an impact on sleep behavior in this population [66].

Further clues may also come from the correlation with brain imaging and with the pattern of brain lesions underlying CP; only a few studies have examined how specific brain damage or hypoxic events that are common causes of CP can affect sleep architecture [6, 8], such as how a dopaminergic dysfunction or a lesion of the basal ganglia, typical of the dyskinetic form of CP, could lead to motor restlessness during sleep that could account for hyperkinesia, hypnic jerks, rhythmic movement disorders, and bruxism [24, 25]. Research into this neglected field is needed in order to implement appropriate and efficient interventions which will allow children with CP and their families to get back to sleep.

### Practice Points for Sleep Disorders in Children with CP

- Prevalence: 19–63% according to different studies
- Etiology: Primary motor impairment, visual impairment, epilepsy and antiepileptic drugs, severity of motor impairment, and psychiatric problems
- Diagnosis: Questionnaires (SDSC) in routine clinical practice to screen for sleep disorders; PSG as needed; and actigraphy in research studies
- Treatment: Parent-based education about sleep and behavioral interventions, melatonin (0.5–5 mg)

## References

- Himmelman K, Uvebrant P. The panorama of cerebral palsy in Sweden. XI. Changing patterns in the birth-year period 2003–2006. *Acta Paediatr.* 2014;103:618–24.
- The definition and classification of cerebral palsy. *Dev Med Child Neurol Suppl.* 2007;109:1–44.
- Angriman M, Caravale B, Novelli L, Ferri R, Bruni O. Sleep in children with neurodevelopmental disabilities. *Neuropediatrics.* 2015;46:199–210.
- Simard-Tremblay E, Constantin E, Gruber R, Brouillette RT, Shevell M. Sleep in children with cerebral palsy: a review. *J Child Neurol.* 2011;26:1303–10.
- Newman CJ, O'Regan M, Hensey O. Sleep disorders in children with cerebral palsy. *Dev Med Child Neurol.* 2006;48:564–8.
- Romeo DM, Brogna C, Quintiliani M, Baranello G, Pagliano E, Casalino T, et al. Sleep disorders in children with cerebral palsy: neurodevelopmental and behavioral correlates. *Sleep Med.* 2014;15:213–8.
- Romeo DM, Brogna C, Musto E, Baranello G, Pagliano E, Casalino T, et al. Sleep disturbances in preschool age children with cerebral palsy: a questionnaire study. *Sleep Med.* 2014;15:1089–93.
- Hayashi M, Inoue Y, Iwakawz Y, Sasaki H. REM sleep abnormalities in severe athetoid cerebral palsy. *Brain Dev.* 1990;12:494–7.
- Kotagal S, Gibbons VP, Stith JA. Sleep abnormalities in patients with severe cerebral palsy. *Dev Med Child Neurol.* 1994;36:304–11.
- Sandella DE, O'Brien LM, Shank LK, Warschausky SA. Sleep and quality of life in children with cerebral palsy. *Sleep Med.* 2011;12:252–6.
- Wayte S, McCaughey E, Holley S, Annaz D, Hill CM. Sleep problems in children with cerebral palsy and their relationship with maternal sleep and depression. *Acta Paediatr.* 2012;101:618–23.
- Ong LC, Yang WW, Wong SW, al Siddiq F, Khu YS. Sleep habits and disturbances in Malaysian children with cerebral palsy. *J Paediatr Child Health.* 2010;46:80–4.
- Mol EM, Monbaliu E, Ven M, Vergote M, Prinzie P. The use of night orthoses in cerebral palsy treatment: sleep disturbance in children and parental burden or not? *Res Dev Disabil.* 2012;33:341–9.
- Didden R, Sigafos J. A review of the nature and treatment of sleep disorders in individuals with developmental disabilities. *Res Dev Disabil.* 2001;22:255–72.
- Galland BC, Elder DE, Taylor BJ. Interventions with a sleep outcome for children with cerebral palsy or a post-traumatic brain injury: a systematic review. *Sleep Med Rev.* 2012;16:561–73.
- Shibagaki M, Kiyono S, Takeuchi T. Nocturnal sleep in mentally retarded infants with cerebral palsy. *Electroencephalogr Clin Neurophysiol.* 1985;61:465–71.
- Hsiao KH, Nixon GM. The effect of treatment of obstructive sleep apnea on quality of life in children with cerebral palsy. *Res Dev Disabil.* 2008;29:133–40.
- McCarthy SF, Gaebler-Spira D, Harvey RL. Improvement of sleep apnea in a patient with cerebral palsy. *Am J Phys Med Rehabil.* 2001;80:540–2.
- Elsayed RM, Hasanein BM, Sayyah HE, El-Auoty MM, Tharwat N, Belal TM. Sleep assessment of children with cerebral palsy: using validated sleep questionnaire. *Ann Indian Acad Neurol.* 2013;16:62–5.
- Zarowski M, Mojs E, Gajewska E, Steinborn B, Samborski W. Prevalence of sleep problems in children with cerebral palsy. *Ann Acad Med Stetin.* 2008;54:59–64.
- Shintani T, Asakura K, Ishi K, Yoshida M, Kataura A, Ogasawara H. Obstructive sleep apnea in children with cerebral palsy. *Nippon Jibiinkoka Gakkai Kaiho.* 1998;101:266–71.
- Stores G. Sleep disturbance in childhood epilepsy: clinical implications, assessment and treatment. *Arch Dis Child.* 2013;98:548–51.
- Schmitt B. Sleep and epilepsy syndromes. *Neuropediatrics.* 2015;46:171–80.
- Lazarus M, Huang ZL, Lu J, Urade Y, Chen JF. How do the basal ganglia regulate sleep-wake behavior? *Trends Neurosci.* 2012;35:723–32.
- Pereira AM, Bruni O, Ferri R, Palmi A, Nunes ML. The impact of epilepsy on sleep architecture during childhood. *Epilepsia.* 2012;53:1519–25.
- Romeo DM, Cioni M, Distefano A, Battaglia LR, Costanzo L, Ricci D, De Sanctis R, Romeo MG, Mazzone D, Mercuri E. Quality of life in parents of children with cerebral palsy: is it influenced by the child's behaviour? *Neuropediatrics.* 2010;41:121–6.
- McDermott S, Coker AL, Mani S, et al. A population-based analysis of behaviour problems in children with cerebral palsy. *J Pediatr Psychol.* 1996;21:447–63.
- Paavonen EJ, Porkka-Heiskanen T, Lahikainen AR. Sleep quality, duration and behavioral symptoms among 5–6-year-old children. *Eur Child Adolesc Psychiatry.* 2009;18:747–54.
- Newman CJ. Sleep: the other life of children with cerebral palsy. *Dev Med Child Neurol.* 2014;56:610–1.
- Atmawidjaja RW, Wong SW, Yang WW, Ong LC. Sleep disturbances in Malaysian children with cerebral palsy. *Dev Med Child Neurol.* 2014;56:681–5.

31. Masters IB, Harvey JM, Wales PD, O'Callaghan MJ, Harris MA. Clinical versus polysomnographic profiles in children with obstructive sleep apnoea. *J Paediatr Child Health*. 1999;35:49–54.
32. Mouthon AL, Huber R. Methods in pediatric sleep research and sleep medicine. *Neuropediatrics*. 2015;46:159–70.
33. Iber C, Ancoli-Israel S, Chesson AL, Quan SF. The AASM manual for the scoring of sleep and associated events: rules, terminology and technical specifications. 1st ed. Westchester: American Academy of Sleep Medicine; 2007.
34. Hill CM, Parker RC, Allen P, Paul A, Padoa KA. Sleep quality and respiratory function in children with severe cerebral palsy using night-time postural equipment: a pilot study. *Acta Paediatr*. 2009;98:1809–14.
35. Hartzell LD, Guillory RM, Munson PD, Dunham AK, Bower CM, Richter GT. Tongue base suspension in children with cerebral palsy and obstructive sleep apnea. *Int J Pediatr Otorhinolaryngol*. 2013;77:534–7.
36. Giannasi LC, Matsui MY, de Freitas Batista SR, Hardt CT, Gomes CP, et al. Effects of neuromuscular electrical stimulation, laser therapy and LED therapy on the masticatory system and the impact on sleep variables in cerebral palsy patients: a randomized, five arms clinical trial. *BMC Musculoskelet Disord*. 2012;13:71.
37. O'Neil ME, Fragala-Pinkham MA, Forman JL, Trost SG. Measuring reliability and validity of the ActiGraph GT3X accelerometer for children with cerebral palsy: a feasibility study. *J Pediatr Rehabil Med*. 2014;7:233–40.
38. Shibagaki M, Kiyono S, Takeuchi T. Delta and spindle components in compressed spectral array during nocturnal sleep in infants with cerebral palsy. *Electroencephalogr Clin Neurophysiol*. 1986;64:493–500.
39. Chervin RD, Hedger K, Dillon JE, Pituch KJ. Pediatric Sleep Questionnaire (PSQ): validity and reliability of scales for sleep-disordered breathing, snoring, sleepiness, and behavioral problems. *Sleep Med*. 2000;1:21–32.
40. Owens JA, Spirito A, McGuinn M. The Children's Sleep Habits Questionnaire (CSHQ): psychometric properties of a survey instrument for school-aged children. *Sleep*. 2000;23:1043–51.
41. Bruni O, Ottaviano S, Guidetti V, et al. The Sleep Disturbance Scale for Children (SDSC). Construction and validation of an instrument to evaluate sleep disturbance in childhood and adolescence. *J Sleep Res*. 1996;5:251–61.
42. Romeo DM, Bruni O, Brogna C, Ferri R, Galluccio C, De Clemente V, et al. Application of the Sleep Disturbance Scale for Children (SDSC) in preschool age. *Eur J Paediatr Neurol*. 2013;17:374–82.
43. Hoban TF. Sleep disorders in children. *Ann N Y Acad Sci*. 2010;1184:1–14.
44. Grigg-Damberger M, Ralls F. Treatment strategies for complex behavioral insomnia in children with neurodevelopmental disorders. *Curr Opin Pulm Med*. 2013;19:616–25.
45. Wyatt K, Edwards V, Franck L, Britten N, Creanor S, Maddick A, Logan S. Cranial osteopathy for children with cerebral palsy: a randomised controlled trial. *Arch Dis Child*. 2011;96:505–12.
46. Duncan B, Barton L, Edmonds D, Blashill BM. Parental perceptions of the therapeutic effect from osteopathic manipulation or acupuncture in children with spastic cerebral palsy. *Clin Pediatr (Phila)*. 2004;43:349–53.
47. Hernandez-Reif M, Field T, Largie S, Diego M, Manigat N, Seoanes M, et al. Cerebral palsy symptoms in children decreased following massage therapy. *Early Child Dev Care*. 2005;175:445–56.
48. Beider S, Moyer CA. Randomized controlled trials of pediatric massage: a review. *Evid Based Complement Alternat Med*. 2007;4:23–34.
49. Glew GM, Fan MY, Hagland S, Bjornson K, Beider S, McLaughlin JF. Survey of the use of massage for children with cerebral palsy. *Int J Ther Massage Bodywork*. 2010;3:10–5.
50. Barlow JH, Powell LA, Gilchrist M, Fotiadou M. The effectiveness of the Training and Support Program for parents of children with disabilities: a randomized controlled trial. *J Psychosom Res*. 2008;64:55–62.
51. Cohen SR, Lefavre JF, Burstein FD, Simms C, Kattos AV, Scott PH, et al. Surgical treatment of obstructive sleep apnea in neurologically compromised patients. *Plast Reconstr Surg*. 1997;99:638–46.
52. Magardino TM, Tom LW. Surgical management of obstructive sleep apnea in children with cerebral palsy. *Laryngoscope*. 1999;109:1611–5.
53. Kontorinis G, Thevasagayam MS, Bateman ND. Airway obstruction in children with cerebral palsy: need for tracheostomy? *Int J Pediatr Otorhinolaryngol*. 2013;77:1647–50.
54. Ramstad K, Jahnsen R, Loftod B, Skjeldal OH. Continuous intrathecal baclofen therapy in children with cerebral palsy – when does improvement emerge? *Acta Paediatr*. 2010;99:1661–5.
55. Morton RE, Gray N, Vloeberghs M. Controlled study of the effects of continuous intrathecal baclofen infusion in non-ambulant children with cerebral palsy. *Dev Med Child Neurol*. 2011;53:736–41.
56. Lundy CT, Doherty GM, Fairhurst CB. Botulinum toxin type A injections can be an effective treatment for pain in children with hip spasms and cerebral palsy. *Dev Med Child Neurol*. 2009;51:705–10.
57. Rios ER, Venâncio ET, Rocha NF, Woods DJ, Vasconcelos S, Macedo D, et al. Melatonin: pharmacological aspects and clinical trends. *Int J Neurosci*. 2010;120:583–90.
58. Wasdell MB, Jan JE, Bomben MM, Freeman RD, Rietveld WJ, Tai J, et al. A randomized, placebo-controlled trial of controlled release melatonin treatment of delayed sleep phase syndrome and impaired sleep maintenance in children with neurodevelopmental disabilities. *J Pineal Res*. 2008;44:57–64.
59. Dodge NN, Wilson GA. Melatonin for treatment of sleep disorders in children with developmental disabilities. *J Child Neurol*. 2001;16:581–4.
60. Ross C, Davies P, Whitehouse W. Melatonin treatment for sleep disorders in children with neurodevelopmental disorders: an observational study. *Dev Med Child Neurol*. 2002;44:339–44.
61. Appleton RE, Jones AP, Gamble C, Williamson PR, Wiggs L, Montgomery P, et al. The use of melatonin in children with neurodevelopmental disorders and impaired sleep: a randomised, double-blind, placebo-controlled, parallel study (MENDS). *Health Technol Assess*. 2012;16:i-239.
62. Sheldon SH. Pro-convulsant effects of oral melatonin in neurologically disabled children. *Lancet*. 1998;351:1254.
63. Schwichtenberg AJ, Malow BA. Melatonin treatment in children with developmental disabilities. *Sleep Med Clin*. 2015;10:181–7.
64. Lo HS, Yang CM, Lo HG, Lee CY, Ting H, Tzang BS. Treatment effects of gabapentin for primary insomnia. *Clin Neuropharmacol*. 2010;33:84–90.
65. Robinson AA, Malow BA. Gabapentin shows promise in treating refractory insomnia in children. *J Child Neurol*. 2013;28:1618–21.
66. Romeo DM, Olivieri G, Brogna C. Use of melatonin for sleep disorders in children with cerebral palsy. *Brain Disord Ther*. 2015;4:5. <https://doi.org/10.4172/2168-975X.1000201>.