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Sleep problems affect 30–55% of typically developing children [1–4] and up to 95% of children with neurodevelopmental disorder (NDD) [5–7]. Children with NDD such as attention deficit hyperactivity disorder (ADHD), autism spectrum disorder (ASD), fetal alcohol spectrum disorder (FASD), and cerebral palsy (CP) have sleep disturbances similar to children without NDD, with the most frequent cause of sleep disturbance in children with and without NDD being behavioral insomnia (behaviors that impact on sleep, including difficulties in initiating or maintaining sleep) [8]. There are, however, several factors particularly associated with NDD that can disturb sleep, such as pain, reflux, seizures, disordered breathing, and restless leg syndrome [9].

This chapter will highlight the characteristics of CP in children, sleep disturbances that are particularly prevalent in children with CP, diagnostic measures, and treatment/interventions, as well as impact of sleep problems on children with CP and their families.

Cerebral Palsy: The Most Common Cause of Physical Disability Among Children Worldwide

One of the NDDs that is associated with sleep problems is cerebral palsy (CP), the most common cause of physical disability among children worldwide [10, 11] with a prevalence of 1.5–2.5 per 1000 live births in developed countries [12–14]. CP is a group of disorders of movement, motor develop-

ment, and posture attributed to nonprogressive central nervous system abnormalities that occur in the developing fetal or infant brain [15, 16]. These motor delays cause limitations in activity, are often associated with abnormal muscle tone, contractures, and deformities [17], and are often accompanied by disturbances of sensation, cognition, communication, perception, behavior, and by epilepsy [11, 15].

CP is classified using the Gross Motor Function Classification System (GMFCS), differentiating severity of mobility and ambulation: GMFCS I, ambulatory; II, walks without aids; III, walks with aids; IV, mobility requires wheelchair or assistance; and V, dependent for mobility [18] (Fig. 58.1). Children with GMFCS IV and V often have the most severe form of CP. There are also several subtypes of CP, namely, spastic diplegia, hemiplegia, and quadriplegia (depending on number and type of limb involvement) as well as dyskinesia, hypotonia, ataxic, and mixed subtypes [16] (Fig. 58.2). Children with spastic quadriplegia are often children with severe motor abnormalities and comorbidities.

Associated medical conditions in children with CP are quite common and can hinder sleep in children with CP. The main comorbidities are sleep-disordered breathing (SDB), seizure disorders, visual impairments, circadian rhythm abnormalities, motor impairments leading to pain and other disturbances, and gastrointestinal abnormalities (related to pain, gastroesophageal reflux, disturbances from gastric tube feedings), drooling and uncoordinated swallowing, intellectual disabilities, and psychological factors [17, 19–22]. In the next section, the prevalence of sleep disorders in children with CP will be described, and the association of these comorbidities and risk of sleep problems will be elaborated.

Prevalence of Sleep Disorders in Children with CP

The prevalence of sleep disorders in children in CP is quite variable and depends on several factors, including GMFCS, CP subtype, age (e.g., preschool vs school aged), and comor-

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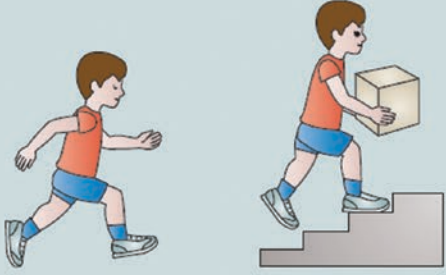
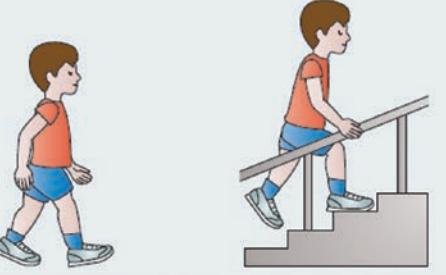

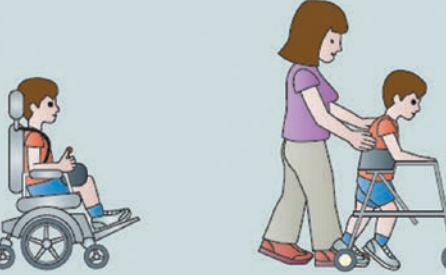
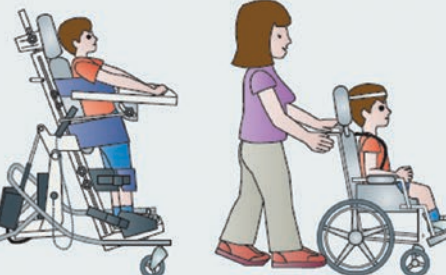
GMFCS expanded and revised between 6 th and 12 th birthday: descriptors and illustrations	
	<p>GMFCS level I Children walk at home, school, outdoors and in the community. They can climb stairs without the use of a railing. Children perform gross motor skills such as running and jumping, but speed, balance and coordination are limited.</p>
	<p>GMFCS level II Children walk in most settings and climb stairs holding onto a railing. They may experience difficulty walking long distances and balancing on uneven terrain, inclines, in crowded areas or confined spaces. Children may walk with physical assistance, a hand-held mobility device or use wheeled mobility over long distances. Children have only minimal ability to perform gross motor skills such as running and jumping.</p>
	<p>GMFCS level III Children walk using a hand-held mobility device in most indoor settings. They may climb stairs holding onto a railing with supervision or assistance. Children use wheeled mobility when travelling long distances and may self-propel for shorter distances.</p>
	<p>GMFCS level IV Children use methods of mobility that require physical assistance or powered mobility in most settings. They may walk for short distances at home with physical assistance or use powered mobility or a body support walker when positioned. At school, outdoors and in the community children are transported in a manual wheelchair or use powered mobility.</p>
	<p>GMFCS level V Children are transported in a manual wheelchair in all settings. Children are limited in their ability to maintain antigravity head and trunk postures and control leg and arm movements.</p>

Fig. 58.1 The Gross Motor Function Classification System (GMFCS expanded and revised) for children with cerebral palsy, 6–12 years of age. This figure depicts GMFCS levels I–V, classifying severity based

on mobility and ambulation. Children with GMFCS IV and V often have the most severe form of CP. (From Graham et al. [70]. Published 2016 Jan 7. Reprinted with permission from Springer Nature)

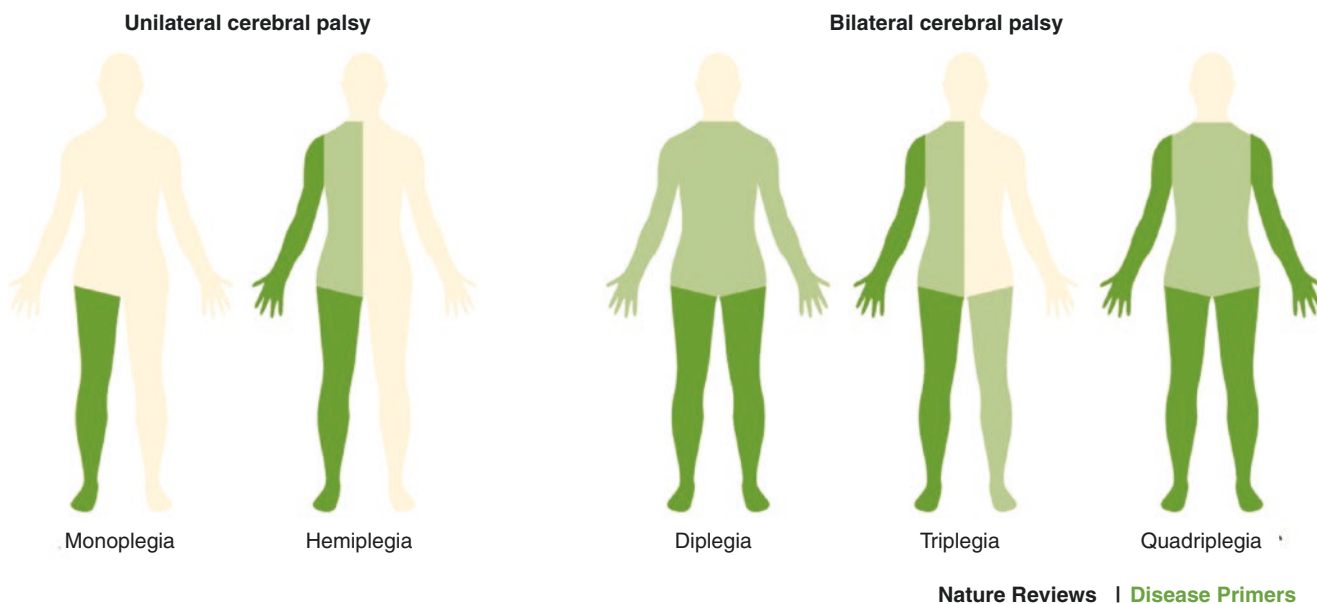


Fig. 58.2 Topographical representation of cerebral palsy. This picture shows the different subtypes of CP: unilateral (monoplegia, hemiplegia) and bilateral (diplegia, triplegia, quadriplegia). (From Graham et al. [70]. Published 2016 Jan 7. Reprinted with permission from Springer Nature)

bilities. Prevalence rates of sleep problems in children with CP have been reported to be as low as 13% to as high as 46% [17, 21–23].

In a recent systematic review and meta-analysis assessing prevalence of sleep disorders in children with CP, we found that the prevalence of sleep disorders as measured by an abnormal total score on the Sleep Disturbance Scale for Children, SDSC, ranges from 13% to 36%, with a pooled prevalence of 23.4% (95% CI 18.8–28.4%). We also found that behavioral insomnia and disorders of initiation and maintenance of sleep (DIMS) were most prevalent of all sleep disorders reported by parents and caregivers. Notably, several studies also showed high prevalence of other sleep disturbances, including sleep breathing disorders, parasomnias (such as bruxism, nightmares), and restless legs. Daytime symptoms such as excessive daytime sleepiness were also highly prevalent (ranging from 12.8% to 63.5%).

A few studies have shown that school-aged children have a higher prevalence of sleep problems as compared to preschool-aged children [22, 24, 25]. Moreover, children with more severe CP (by GMFCS or by CP subtype) have higher prevalence of sleep disturbances [24–26].

Types of Sleep Disorders in Children with CP

Children with CP are at risk of several types of sleep disorders. Not surprisingly, many sleep disorders in children with CP are similar to those present in children with other NDDs and in typically developing children. Children with CP are at risk of sleep disturbances, including behavioral

insomnia, DIMS, sleep-disordered breathing, and sleep disruption secondary to pain and problems with sensory processing and/or associated medical conditions (motor impairment/muscle spasms, seizures, gastroesophageal reflux [GERD], and feeding issues) [17, 21–23, 24, 25]. One study showed that ~80–90% of children with severe CP have a chronic gastrointestinal condition, with GERD being most common [17]. Children with CP taking medications for comorbid medical conditions may also have side effects affecting sleep.

Notably, some children with CP also have multiple and concurrent sleep disorders. Romeo and colleagues found that 42% of their study population of children with CP had at least one sleep disorder (with an abnormal score on at least one SDSC factor) [24]. Similarly, in our study of Canadian children with CP, a high percentage (44%) of children in our cohort had one or more sleep disorder [22]. Interestingly, we also found that school-aged children had a higher prevalence of multiple sleep disorders than preschool-aged children (59.5% vs 24.2%, respectively).

Behavioral Insomnia

The most common sleep disorder in children with CP is behavioral insomnia. The prevalence of behavioral insomnia in children with CP ranges from 36% to 89% [27–29]. The findings from our systematic review and meta-analysis on sleep problems in children with CP demonstrated that as per the SDSC, the most common sleep disorder in children with CP was DIMS (12–50%) [27, 30, 31], followed by sleep-

wake transition disorders, sleep breathing disorders, disorders of arousal, disorders of excessive somnolence, and lastly, sleep hyperhidrosis.

Sleep-Disordered Breathing

Compared to typically developing children, children with CP are at higher risk of SDB (from snoring to the most severe form on the SDB spectrum, obstructive sleep apnea) [19, 21, 32–34]. The prevalence of SDB among children with CP ranges from 12% to 79% [19, 24, 25, 27, 28, 30, 35]. Snoring is common in children with CP (37–78%) with higher rates compared to controls [27, 35]. CP places children at higher risk of SDB due to a number of factors, including craniofacial abnormalities specific to children with CP (including maxillary hypoplasia, palatal hypotonia, glossoptosis, retrognathia, laryngomalacia, and laryngeal dystonia), abnormal tone of upper airway muscles, and primary central control of breathing abnormalities [23, 36–38]. However, it is important to note that children with CP may also have SDB secondary to the common causes similar to typically developing children, such as adenotonsillar hypertrophy, nasal/allergic rhinitis and inflamed/enlarged turbinates, and obesity. Often the etiology of SDB in children with CP is multi-factorial.

Other Sleep Disorders

Similar to children without CP, children with CP can have parasomnias, such as nightmares, sleep talking, and sleepwalking [21, 22]. Of particular note, a few studies showed a high prevalence of bruxism in children with CP, ranging from 23% to 38% [28, 39]. Higher rates of bruxism are reported in preschool-aged children with CP compared to school-aged children [28]. Restless legs and restless leg syndrome are also prevalent in children with CP [27, 28].

Medical conditions are common in children with CP and can be associated with or lead to sleep problems. These medical conditions include motor impairment and muscle spasms, seizure disorder/epilepsy, gastrointestinal issues, auditory and visual problems, pain, and consequences of treatment of medical conditions related to CP, such as medications or orthotic devices.

Impact of Sleep Problems on Children with CP and Their Families

Negative effects of sleep disorders on children with CP may include both daytime and sleep symptoms, including daytime sleepiness, napping or falling asleep during the day, and poor daytime functioning, which may lead to decreased par-

ticipation in daytime activities, behavior issues (irritability, hyperactivity, impulsivity) and poor quality of life [40–42]. One study found that this interplay of poor quality of life, sleep issues, and pain also is prevalent in young adults with CP [43].

Children with CP have neurodevelopmental and behavioral issues. One study in school-aged children with CP showed a moderate correlation between having a sleep disorder (as measured by the total SDSC score) and the child's behavior (as measured by the Child Behavior Checklist (CBCL)) [24]. A knowledge gap in the CP literature was the association between sleep problems and behavior issues in younger children and the interplay between sleep problems, behavior issues, and pain. We conducted a study to help fill this knowledge gap and showed for the first time the association between *specific* sleep problems and behavioral difficulties in children with CP, finding that several sleep problems (including sleep anxiety, difficulty getting to sleep, and frequent awakenings) were associated with behavioral difficulties [44]. We found that peer problems were the most common behavioral difficulty, while emotional symptoms and hyperactivity were also highly prevalent. There were differences between preschool-aged and school-aged children, with the prevalence of behavioral issues being 17.6% and 29.1% of preschool- and school-aged children, respectively; specifically, the prevalence of peer problems was 23.5% and 30.4%, respectively [44].

As sleep problems lead to negative effects on children with CP, sleep problems also can negatively impact the families/caregivers [45]. In particular, sleep problems in children with CP may lead to sleep deprivation and sleep loss in the family/caregivers due to sleep disturbances from frequent nighttime awakenings from observing/monitoring their child's sleep or feedings. The combination of these issues may lead to parental/caregiver sleep disturbances that, in turn, may impact quality of life of family. Compared to typically developing children, children with CP have poorer health-related quality of life (HRQoL) with lower physical, social, school, and emotional functioning [40, 41]. In our cohort of Canadian children with CP, we found that 33% of children have poor overall HRQoL (more than two standard deviations below normative values) and that children with poor HRQoL were more likely to be non-ambulatory, to have sleep problems, and to have significant comorbidity (controlled for age and pain) [42].

Physical Exam and Diagnostic Measures for Sleep Disorders in Children with CP

Physical exam findings for sleep-disordered breathing (SDB)/obstructive sleep apnea (OSA) in children with CP are similar to that of children without CP and involve the fol-

lowing: examination of upper airway, specifically assessment of tonsillar size, nasal turbinates for inflammation, polyps or nasal septal deviation, midfacial hypoplasia, “adenoidal facies” (from chronic mouth breathing secondary to adenoidal hypertrophy), and maxillary and/or mandibular hypoplasia. Children with CP also have risk of palatal hypotonia, glossoptosis, retrognathia, laryngomalacia and laryngeal dystonia, and abnormal tone of upper airway muscles [23, 36–38].

Diagnostic measures for sleep disorders include polysomnography in a sleep laboratory, nocturnal home oximetry, and actigraphy. Laboratory polysomnography is the gold standard diagnostic tool for SDB as well as for periodic leg movement disorder. Nocturnal home oximetry has been used as a screening test for OSA in children [46, 47]; though not specifically validated for children with CP, nocturnal oximetry can be useful in detecting desaturations in children with CP and can potentially help with screening and prioritization for laboratory polysomnography. Actigraphy, a wristband worn to assess movement (proxy for wake) vs non-movement (proxy for sleep) is used to determine sleep patterns, sleep fragmentation, and sleep duration in children; actigraphy has been used to assess sleep in children with NDD such as CP [48–51], though it is likely more challenging in children with CP who are less mobile during sleep (e.g., children with CP with severe impairments, quadriplegia).

Subjective tools such as sleep logs/diaries and sleep questionnaires have been used to assess parental report of sleep problems in children. Specifically, in children with CP, we found that the most common parental questionnaire is the SDSC (sleep disturbance scale for children) [21], a validated questionnaire for children 3–18 years consisting of 26 sleep-related behaviors [52]. In addition to a total SDSC score, there are also four SDSC subscale scores: (1) disorders of initiation and maintenance of sleep, (2) sleep breathing disorders, (3) disorders of excessive somnolence, and (4) sleep hyperhidrosis [52]. Another tool used to assess sleep in children with CP is the Pediatric Sleep Questionnaire (PSQ) [53], a validated tool for children 2–18 years old comprised of items including snoring, sleep-disordered breathing, sleepiness, and behavioral problems.

Sleep Interventions for Children with CP and Sleep Disorders

As in typically developing children and children with NDDs, the main intervention for children with CP and sleep disorders is behavioral sleep interventions [54–56]. These strategies include optimization of sleep habits/sleep “hygiene” and the sleep environment, ensuring a regular and consistent sleep routine, and avoidance or minimization of external stimuli prior to bedtime (e.g., light, noise, screen time). We

are currently in the recruitment phase of our Canadian-wide randomized controlled trial to assess the impact of our online behavioral sleep intervention tool (BetterNightsBetterDays-NDD), which we tailored for families of children with NDD (including CP) [57, 58].

Another strategy that may be helpful is incorporating relaxation techniques or activities (such as reading, music, light touch with soft/dim lighting) as part of the wind-down period prior to bedtime and as part of the bedtime routine. Massage therapy by a qualified massage therapist for children with CP may also help with sleep and relaxation in some children, with one before-after study showing improvements in child’s mobility, eating, and sleep quality, as well as improved parental well-being [59]. Massage therapy may be contraindicated in some children with CP; thus, the decision for massage therapy should be made in conjunction with the child’s medical team.

Treating underlying medical conditions related to CP is important, as many of these comorbid conditions can impact sleep: conditions related to motor impairment of CP, including muscle spasms, spasticity, and inability to move independently in bed. Changing body position during sleep may lead to sleep disruption; however, change in position may also help with discomfort and in prevention of pressure sores, thus decreasing nocturnal pain and potentially improving sleep quality. Similarly, wearing splints or orthotic devices for motor impairments can lead to sleep disturbance from discomfort from these devices; however, often these devices can also help with muscle spasms and spasticity which can lead to improved sleep quality. Pharmacology treatment, such as Baclofen, can alleviate symptoms of hypertonia and spasticity. Studies found that in children taking Baclofen, there was parental report of improvement of the child’s spasticity during sleep and reduction of nighttime awakenings [60, 61]. Children with CP have a high prevalence of seizure disorder which may lead to sleep disturbance and nighttime awakenings and ultimately daytime sleepiness. Treating the underlying seizure disorder and ensuring good seizure control can help with overall sleep and thus daytime functioning.

In children with CP and SDB/OSA, treatment of the underlying cause can help with sleep quality and symptoms of OSA [62–64]. If nasal obstruction secondary to allergic rhinitis and/or adenoidal hypertrophy are present, then medical treatment with nasal steroids (if no contraindications) may help decrease inflammation and may help reduce adenoidal size. Referral to specialists may help for management if child is a surgical candidate for adenoidectomy and/or tonsillectomy or a candidate for noninvasive ventilation. Treating the upper airway obstruction with medical or surgical treatment may help with quality of sleep with improvement of restless sleep/awakenings from these breathing disturbances during sleep.

Children with CP may also have difficulty swallowing, and/or pain from gastrointestinal reflux, and some children may have gastric feeding. Reflux and nighttime feedings can disturb sleep. Treatment of underlying reflux can help improve sleep symptoms; moreover, timing of feeds can sometimes be scheduled to minimize sleep disruption.

Sleep positioning systems are sometimes used in children with CP during sleep to help prevent hip migration, especially those children with CP who are non-ambulatory. A recent Cochrane review assessed the effectiveness of these sleep positioning systems on reduction or prevention of hip migration as well as the impact on pain reduction during sleep and on sleep quality [65]. There were very few studies in the literature, and the review did not show significant differences on hip migration, pain, and sleep quality in children using and not using these sleep positioning systems [65].

In children with NDDs, limited research has shown effectiveness of melatonin in treating sleep problems in children [66, 67]. Specifically, studies have shown improvement in sleep latency and awakenings; however, the impact on sleep duration and sleep quality remains unclear. Of note, no studies have been done specifically assessing the effectiveness of melatonin on children with CP [56, 68]. Moreover, the long-term safety and efficacy/effectiveness of melatonin in children with and without NDDs have not been determined. Despite these facts, melatonin is commonly used in children with NDDs; in our Canadian cohort of children with CP, we found that 21% of children had used melatonin for sleep [22]. As suggested in a national review on melatonin and sleep disorders in children (Canadian Paediatric Society), more robust studies in children, in particular randomized controlled trials, are required to determine the clinical effectiveness on sleep outcomes and safety of melatonin in children [69]. Notably, in light of the high prevalence of sleep disorders in children with CP and other NDDs, effectiveness studies are critically needed for this at-risk population.

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