185 Nonobstructive Pediatric Sleep Disorders

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KEY POINTS

- Although most otolaryngologists are very familiar with pediatric obstructive sleep apnea, nonobstructive sleep disorders are also very common causes of morbidity in children.
- A *dyssomnia* is a primary sleep disorder characterized by an abnormality in the amount, quality, or timing of sleep that results in difficulty in initiating or maintaining sleep.
- A *parasomnia* is an undesirable event that occurs during entry into sleep, within sleep, or during arousals from sleep; parasomnias are broadly characterized by occurrence during rapid-eye-movement (REM) or non-REM sleep.
- Important pediatric dyssomnia conditions include insufficient sleep syndrome, narcolepsy, sleep-related movement disorders, circadian sleep-wake disorders, idiopathic hypersomnia, Kleine-Levin syndrome, and chronic insomnia disorder.
- Insufficient sleep duration is by far the most common cause of sleepiness in children and adolescents; it may coincide with chronic insomnia disorder and/or delayed sleep-wake phase disorder.
- Non-REM pediatric parasomnias include sleep terrors, sleepwalking, confusional arousals, and other events occurring during slow-wave sleep.
- Pediatric REM parasomnias occur later in the night and include nightmare disorder, recurrent isolated sleep paralysis, and REM sleep behavior disorder.
- Sleep disruption is commonly associated with neuropsychiatric disorders such as attention deficit hyperactivity disorder and autism spectrum disorder, either as a primary manifestation or as a side effect of pharmacologic treatment.

Although the clinical significance of pediatric obstructive sleep apnea syndrome is appropriately recognized, nonobstructive pediatric sleep disorders may be lesser known but still very common major causes of morbidity for children and their families. Disturbances in the child's normal sleep architecture from any source may significantly affect the child's development and emotional and behavioral well-being. Multiple factors including nighttime awakenings, sleep latency, longest sleep period, and daily naps affect how a caretaker perceives the quality of sleep a child is achieving nightly.[1] These features of sleep will naturally change in healthy children as they get older; however, progression of these factors may result in nonobstructive pediatric sleep disorders. The *International Classification of Sleep Disorders*, third edition (ICSD-3), provides a framework for the nomenclature, characterization, and diagnosis of these disorders, which can be categorized as either dyssomnias or parasomnias.[2] Although not categorized directly as a dyssomnia or parasomnia, neuropsychiatric disorders such as attention deficit hyperactivity disorder (ADHD) or pervasive developmental disorders have also been regularly associated with sleep disturbance in pediatric patients.

DYSSOMNIAS

The *Diagnostic and Statistical Manual of Mental Disorders*, fifth edition (DSM-V), defines a dyssomnia as a primary disorder of sleep or wakefulness characterized by insomnia or hypersomnia as the major presenting symptom. Dyssomnias are disorders of the amount, quality, or timing of sleep.[3]

Insufficient Sleep Syndrome

Insufficient sleep is a very common cause of daytime sleepiness in children, particularly adolescents. Many children fall short of getting the recommended amount of sleep per 24 hours, which includes naps (Table 185.1).[4] Sleep duration decreases for a variety of reasons as children age, including later bedtimes as a result of watching television or doing homework.[5] In addition, early school start times may contribute to insufficient sleep.[6] The ramifications of insufficient sleep can include behavioral changes, chronic fatigue, missing school, poor school performance, irritability, and driving accidents.[5] In one prospective study, children who experienced insufficient sleep in preschool and the early school years displayed poorer maternal- and teacher-reported executive function, behavior, and social-emotional function at age 7 than those who slept the appropriate amount.[6] Insufficient sleep can also increase the risk of hypertension, obesity, diabetes, and depression as well as suicidal thoughts in teenagers.[7] Consequently the American Academy of Sleep Medicine has recently proposed that school days for middle and high schoolers should begin at 8:30 a.m. or later in order to promote safe driving, overall health, and better academic performance.[8]

Unfortunately it is quite possible that parents may actually overestimate the amount of sleep their children are getting, as the time of actual sleep onset may be later than the reported bedtime.[6] With this in mind, parents must be vigilant for signs of insufficient sleep syndrome, such as the need to be awakened for school, attempts by the child to catch up on sleep over the weekend, falling asleep at inappropriate times, or behavioral changes.[5] Treatment involves behavioral modifications for both the adolescent and the family, especially if family members subscribe to similar patterns, to reinforce good sleep hygiene for the entire household.

Narcolepsy

Narcolepsy is a rapid-eye-movement (REM) sleep disorder that often starts in adolescence and is classically defined as a combination of irresistible daytime somnolence, sleep onset paralysis, and hypnagogic hallucinations, which are vivid hallucinations on sleep onset.[9] The condition affects approximately .02% to .05% of the US population, but only about 50% of individuals will exhibit the classic triad of symptoms.[10,11] The ICSD-3 has further subdivided narcolepsy into types 1 and 2. Narcolepsy type 1 is known to be caused by a deficiency of the neurotransmitters hypocretin-1 and -2 (orexin-A and -B), which promote wakefulness and prevent inappropriate transition to non-REM or REM sleep. Narcolepsy

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Abstract

Although most otolaryngologists are very familiar with pediatric obstructive sleep apnea, nonobstructive sleep disorders are also very common causes of morbidity in children. A dyssomnia is a primary sleep disorder characterized by an abnormality in the amount, quality, or timing of sleep that results in difficulty in initiating or maintaining sleep. Important pediatric dyssomnia conditions include insufficient sleep syndrome, narcolepsy, sleep-related movement disorders, circadian sleep-wake disorders, idiopathic hypersomnia, Kleine-Levin syndrome, and chronic insomnia disorder. A parasomnia is an undesirable event that occurs during entry into sleep, within sleep, or during arousals from sleep. Parasomnias are broadly characterized by occurrence during rapid-eye-movement (REM) sleep or non-REM sleep. Non-REM pediatric parasomnias include sleep terrors, sleepwalking, confusional arousals, and other events occurring during slow-wave sleep. Pediatric REM parasomnias occur later in the night and include nightmare disorder, recurrent isolated sleep paralysis, and REM sleep behavior disorder. Sleep disruption is commonly associated with neuropsychiatric disorders such as attention deficit hyperactivity disorder (ADHD) and autism spectrum disorder (ASD), either as a primary manifestation or as a side effect of pharmacologic treatment. Providers who evaluate children with sleep problems should have a working knowledge of nonobstructive sleep disorders to complement their expertise in obstructive sleep disorders.

Keywords

Dyssomnia
parasomnia
narcolepsy
hypersomnia
insomnia
restless leg syndrome
insufficient sleep syndrome

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TABLE 185.1 Recommended Sleep Duration on a Regular Basis to Promote Optimal Child Development

Age	Hours
4–12 months	12–16 h/24 h period (including naps)
1–2 years	11–14 h
3–5 years	10–13 h
6–12 years	9–12 h
13–18 years	8–10 h

type 2 is associated with normal hypocretin-1 levels and is of unknown cause.[11,12] Patients with narcolepsy type 1 may also display cataplexy, the sudden but brief loss of muscle tone associated with emotional triggers.[11] Evaluation of suspected narcolepsy should include overnight polysomnography (PSG) data to rule out other sleep disorders, followed by a daytime multiple-sleep latency test (MSLT) or analysis of cerebrospinal fluid for hypocretin-1 deficiency.[11] The MSLT is an objective test that measures the speed of falling asleep during 20-minute napping intervals, with a faster onset of sleep indicating a greater level of sleepiness.[13] A child with narcolepsy will often exhibit spontaneous onset of REM sleep during at least two of these naps, whereas normal subjects will exhibit no REM sleep.[8] The diagnostic criteria for narcolepsy (both types 1 and 2) include a sleep latency of less than 8 minutes on the MSLT, whereas normal sleep latency is approximately 15 minutes.[13] Differentiating between narcolepsy types 1 and 2 relies on the presence of cataplexy and on cerebrospinal fluid hypocretin-1 levels, if measured.[11]

Children with narcolepsy may begin having symptoms as early as 5 years of age, but they may not be diagnosed until much later in adolescence because of confounding emotional and behavioral disturbances.[9,13] Symptoms are often characterized by roughly 10- to 20-minute sleep attacks in which an irresistible urge to sleep invades normal daily activities such as eating, driving, or walking.[10] Cataplexy in addition to intense hypersomnia is pathognomonic for narcolepsy type 1.[10–12] Adequate treatment of children with narcolepsy is important because these children often manifest increased emotional lability, adjustment difficulty, and problems in school.[10] In addition to promoting good sleep hygiene and establishing healthy sleep-wake schedules, treatment includes stimulants to decrease daytime sleepiness, modafinil to promote wakefulness, and suppression of REM sleep with agents such as tricyclic antidepressants or serotonin reuptake inhibitors.[9,13] Additional studies are also looking at agents such as sodium oxybate, which acts as an agonist on the γ-aminobutyric acid (GABA) B receptor, to specifically address the cataplexy component of narcolepsy.[14]

Sleep-Related Movement Disorders

It is important to recognize limb movement disorders such as restless leg syndrome (RLS) and periodic limb movement disorder (PLMD) in children, lest associated pain, sleep disturbances, and possible comorbid neuropsychiatric conditions go untreated. RLS is defined as the subjective complaint of uncomfortable tingling sensations in the lower extremities around bedtime with relief of symptoms with movement, whereas PLMD is the repetitive contraction of the anterior tibialis muscle evaluated objectively in the sleep laboratory (Box 185.1).[13,15] Most patients with RLS will also have periodic limb movements on PSG; however, the converse is not always true.[13] Approximately 2% of the pediatric population surveyed in one study of over 10,000 families met the criteria for a diagnosis of RLS. No gender differences were observed, and 70% reported a family history of RLS.[16–18] Sleep disturbances were common in children with RLS, as was the complaint of "growing pains"; therefore RLS should at least be considered and appropriately treated in children with sleep difficulty and unexplained pain.[16] Renal failure and iron deficiency are two comorbidities that are thought to exacerbate pediatric RLS.[15] Dopaminergic dysfunction results from a deficiency of iron in the central nervous system and aggravates symptoms.[18] When ferritin levels are below 50 μg/L, supplemental iron therapy should at least be considered.[18] Although PLMD may be a benign finding, many children with PLMD exhibit daytime symptoms associated with ADHD and deserve further neuropsychiatric evaluation if clinical suspicion is present.[19] The mainstays of treatment for both RLS and PLMD are dopaminergic therapy, gabapentin, benzodiazepines, and possibly iron therapy.[13,18]

BOX 185.1 Criteria for Diagnosis of Restless Leg Syndrome in Children and Adults

ADULTS AND CHILDREN: ALL FIVE CRITERIA MUST BE MET

1. An urge to move the legs usually but not always accompanied by or felt to be caused by uncomfortable and unpleasant sensations in the legs.[a,b]
2. The urge to move the legs and any accompanying unpleasant sensations begin or worsen during periods of rest or inactivity such as lying down or sitting.
3. The urge to move the legs and any accompanying unpleasant sensations are partially or totally relieved by movement, such as walking or stretching, at least as long as the activity continues.[c]
4. The urge to move the legs and any accompanying unpleasant sensations during rest or inactivity occur only in the evening or night or are worse than during the day.[d]
5. The occurrence of these features is not solely accounted for as symptoms primary to another medical or a behavioral condition (e.g., myalgia, venous stasis, leg edema, arthritis, leg cramps, positional discomfort, habitual foot tapping—otherwise known as "RLS mimics.")

[a]Sometimes the urge to move the legs is present without the uncomfortable sensations and sometimes the arms or other parts of the body are involved in addition to the legs.
[b]For children, the description of these symptoms should be in the child's own words.
[c]When symptoms are very severe, relief by activity may not be noticeable but must have been previously present.
[d]When symptoms are very severe, the worsening in the evening or night may not be noticeable but must have been previously present.
RLS, Restless leg syndrome.
From Picchietti D, Bruni O, de Weerd A, et al: Pediatric restless legs syndrome diagnostic criteria: an update by the International Restless Legs Syndrome Study Group. *Sleep Med* 14:1253–9, 2013.

Sleep-related bruxism is a specific form of sleep-related movement disorder characterized by grinding or clenching of the teeth during sleep, which may result in sleep arousals if severe. The prevalence of bruxism typically decreases with age; however, the persistence of bruxism may be related to separation anxiety in the child and the tendency for the parent to remain in the bedroom until the child falls asleep.[20]

Sleep-related rhythmic movement disorder includes behaviors such as body rocking, head banging, and head rolling, which usually occur at sleep onset and disappear early in childhood.[20,21] Such behavior is typically benign and should be treated primarily by creating a safe environment for the child. If the behavior persists beyond early adolescence, an evaluation for other neuropsychiatric conditions should be pursued.[4]

Circadian Rhythm Sleep-Wake Disorders

Circadian rhythm sleep-wake disorders are quite common in pediatric patients and may not manifest until adolescence, when

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waking for school becomes problematic. Delayed sleep-wake phase syndrome (DSWPS) is a common subset of circadian sleep disorders that affects 5% to 10% of adolescents.[4] The delay of actual sleep 2 to 3 hours beyond the desired bedtime defines DSWPS, and it appears that the ability to initiate sleep does not coordinate with environmental clues such as light and darkness.[4] These children often initiate sleep between 2 and 6 a.m., but the sleep architecture, once initiated, is normal.[13] Prompt treatment of DSWPS with improved sleep hygiene, chronotherapy to incrementally advance bedtime, and melatonin therapy are essential to avoid potential school or family-related conflicts.[4,13]

Idiopathic Hypersomnia and Kleine-Levin Syndrome

Idiopathic hypersomnia is characterized by excessive sleepiness that lasts for at least 4 months.[2,21] When assessed, these children often exhibit normal PSG data.[7] Normal hypocretin levels, the absence of cataplexy, and the lack of REM sleep during prolonged naps distinguish idiopathic hypersomnia from narcolepsy.[13,21] Studies show that stimulant therapy is less effective for hypersomnia than it is for narcolepsy.[13]

Kleine-Levin syndrome is a rare form of hypersomnia that affects adolescents and presents with compulsive hyperphagia and indiscriminate hypersexuality in addition to hypersomnia.[3,9] Other symptoms such as confusion, depression, memory deficits, irritability, and hallucinations can confound the diagnosis. The condition is episodic and self-limited, occurs over a period of weeks to years, and may be associated with completely symptom-free intervals.[21] A 2:1 to 3:1 male to female predominance is apparent, and the median age of onset is 15 years.[3,21] Familial cases are present in roughly 4% of those diagnosed, similar to the range for narcolepsy type 1, and viral infections have been reported as precipitating conditions.[9,21,22] PSGs in patients with Kleine-Levin syndrome demonstrate increased sleep time and short REM sleep latency, consistent with hypersomnia that does not meet the criteria for narcolepsy.[3] Use of modafinil, amphetamines, and methylphenidate has been reported; however, given the extremely sporadic nature of the condition, no consensus on pharmacotherapy exists.[21]

Chronic Insomnia Disorder

Pediatric insomnia comprises the inability to initiate or maintain sleep despite being given the age-appropriate opportunity to sleep.[9,23] The prevalence, although varied, ranges from 1% to 6% in the general population and up to 75% in pediatric patients with comorbid neurocognitive disorders such as ADHD or pervasive developmental disorders.[23] Pediatric insomnia has been attributed to poor sleep hygiene or the inability for a parent to withdraw comfort measures in a fashion that allows the child to self-soothe and initiate sleep; it often presents with paradoxic hyperactivity and restlessness.[9,23] Treatment for insomnia focuses on providing a more consistent routine for the child and enforcing good practices such as caffeine avoidance, limit setting, and an age-appropriate schedule. If a child has persistent issues once the sleep hygiene practices have been addressed, the child should be evaluated for additional neurocognitive comorbidities. Clonidine, mood stabilizers, antidepressants, hypnotics, or antihistamines are commonly prescribed to treat chronic insomnia in patients with neurocognitive disorders based on anecdotal evidence, but there is no consensus on pharmacologic management at this time.[23]

Limit-Setting Sleep Disorder

Limit-setting sleep disorder is a specific, common pediatric behavioral insomnia most often exhibited as a bedtime stalling tactic by the child. This disorder may occur in up to 10% to 30% of toddlers and up to 15% of adolescents[5] and often results from failure of caregivers to set and enforce consistent bedtime routines. The behaviors are often manifested as repeated requests for more bedtime stories, repeated requests to have the parent stay while the child falls asleep, or even fearful behaviors such as crying and clinging. Caregivers may perceive prolonged sleep latency and shorter sleep time; however, the quality of sleep the patient receives once asleep is typically normal.[5] Treatment for limit-setting sleep disorder involves behavioral interventions such as instituting regular and unwavering bedtime routines and promoting good sleep hygiene to reverse the dysfunctional pattern. Many behavioral therapies have demonstrated efficacy in reducing nighttime resistance.[24] The *extinction method* may be one of the most commonly described; it involves ignoring bedtime crying to eventually allow the child to self-soothe and fall asleep without parental intervention. Unfortunately, whereas this method may be quite effective, parental compliance is often not maintained.[25]

PARASOMNIAS

Parasomnias are common in the preschool age group and are defined by the American Academy of Sleep Medicine as an undesirable event that occurs during entry into sleep, within sleep, or during arousals from sleep.[20,26] Etiologic factors include genetic predisposition, disrupted sleep hygiene, associated neuropsychiatric issues such as anxiety or depression, and parental socioeconomic status. These disorders are classified into non-REM and REM types (Table 185.2). The non-REM types occur during deep or slow-wave sleep in the first two-thirds of the night and include partial arousals and motor disturbances.[9] Particular non-REM events include sleep terrors, somnambulism (sleepwalking), confusional arousals, and sleep-related eating disorder. REM parasomnias tend to occur in the last third of the night and include nightmare disorder, sleep paralysis, and REM sleep behavior disorder.[9] Additional parasomnias have been defined that do not fall clearly into a REM or non-REM category, such as somniloquy (sleep talking) and nocturnal enuresis. Although typically benign in adolescence, parasomnias can cause difficulty for both child and family and should be recognized and appropriately addressed. Most parasomnias are self-limited and generally do not occur for more than a few minutes. They are associated with resistance to wakening by the parent and amnesia to the event for the child.[5] Therefore most treatment has focused around supportive care for the child and family and creating a safe sleep environment for the child.

Sleep Terrors

Sleep terrors are described as an arousal from slow-wave sleep that occurs in the first part of the night and may be accompanied by a piercing cry or scream as well as intense fear.[5,20] Sleep terrors

TABLE 185.2 Features of Rapid-Eye-Movement Versus Non-REM Parasomnias

	Rapid Eye Movement (REM)	Non-REM
Types of parasomnia	REM behavior sleep disorder Recurrent sleep paralysis Nightmare disorder	Confusional arousals Sleepwalking Sleep terrors
Time of night	Last third of the night	First two-thirds of the night
Type of sleep	REM sleep	Deep or slow-wave sleep
Recall	Typically vivid recall of events	No recall of events

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often affect children between 3 and 10 years of age and may occur on a weekly or monthly basis.[27] An accompanying autonomic response with tachycardia and tachypnea is usually present. A strong genetic association with sleep terrors has been found.[28] Most symptoms resolve spontaneously over months to years. Prolonged symptoms should be further evaluated to determine whether other common causes of sleep disturbances, such as sleep-disordered breathing, may be contributing to the problem.[27]

Sleepwalking

Somnambulism, more frequently known as *sleepwalking*, is defined as walking around during slow-wave sleep in an altered state of consciousness.[20] Symptoms typically occur during the first third of the night. Although relatively benign, somnambulism represents a safety risk to the child; therefore appropriate precautions should be taken. Additionally, other sleep disorders—such as sleep terrors, RLS, and sleep-disordered breathing—may have to be explored as predisposing or associated factors.[20,26]

Confusional Arousals

Confusional arousals often occur in toddler-aged children and include events such as sitting up in bed and moaning or uttering words such as *no* or *go away* repeatedly.[28] These events may last up to 30 minutes and often occur within 2 to 3 hours of sleep onset.[28,29] They are not associated with an autonomic response and are self-limited, with return to normal sleep on cessation of the event.[28]

Nightmare Disorder

Differing from sleep terrors, nightmare disorder tends to occur exclusively during REM sleep in the earlier hours of the morning as opposed to the first part of the night.[4] The child is usually able to recount the events of a nightmare more easily than in the case of a sleep terror, after which amnesia is quite common. Sometimes return to sleep may be delayed.[29] Treatment is mainly supportive care.

Sleep Paralysis

Sleep paralysis, as the name implies, is the inability to move the body upon falling asleep or awakening. Common in adolescents, the disorder is not associated with daytime sleepiness but does have elements of awareness of surroundings, such as hearing footsteps or sensing the presence of someone nearby.[29] Potential associations with anxiety or even circadian rhythm sleep-wake disorders have been explored. Treatment with behavioral and possibly pharmacologic therapy may be indicated.[28]

Rapid-Eye-Movement Sleep Behavior Disorder

REM sleep behavior disorder is more frequent in adult patients and is quite rare in children, but it may be more common in children with congenital brainstem disturbances.[29] Such disturbances, in which the child exhibits dream-enactment behavior, may be associated with yelling or flailing limbs. The significance of REM sleep behavior disorder in childhood is unknown.[29]

PEDIATRIC NEUROPSYCHIATRIC DISORDERS AND SLEEP DISRUPTION

Neuropsychiatric disorders do not clearly fall into the dyssomnia or parasomnia categories. Common pediatric neuropsychiatric disorders include ADHD, pervasive developmental disorders such as autism spectrum disorders (ASD), and mood disorders such as major depression. Each can lead to significant sleep disturbance in the pediatric patient. It should also be noted that the common pharmacologic treatments for these disorders, such as stimulant therapy for ADHD, frequently affect sleep. It can often be difficult to ascertain clinically whether the sleep disturbance is due to the primary disorder or the side effects of its pharmacologic treatment. Sleep problems can affect up to half of children who have a diagnosis of ADHD.[30] Children with ADHD have a tendency for associated sleep-disordered breathing, limb-movement disorders, circadian rhythm sleep-wake disorders, and narcolepsy. The true impact on daytime functioning for these individuals has yet to be fully explored. However, it is clear that appropriate sleep routines in children with ADHD may be helpful to their overall performance.[4,31] ASDs have been associated with sleep problems in well over 50% of diagnosed individuals.[32] These children, especially those with concurrent intellectual disabilities, display decreased total sleep time and sleep efficiency as well as prolonged sleep onset.[33]

Certainly behavioral therapy, although potentially more difficult in the autistic child, is the most beneficial approach to treat sleep disorders in this population of children. Additionally, melatonin use is effective in decreasing sleep latency and increasing total sleep time in those with autism.[34] Mood disorders such as depression and anxiety may be related to sleep dysfunction such as insomnia, hypersomnolence, and even any of the aforementioned parasomnias.[5,13] Therapy focuses on addressing the underlying cause of the child's depression or anxiety disorder, whether that be through pharmacologic or behavioral therapy.

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