

# Pediatric Obstructive Sleep Apnea Syndrome

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**Objective:** To review evidence-based knowledge of pediatric obstructive sleep apnea syndrome (OSAS).

**Data Sources and Extraction:** We reviewed published articles regarding pediatric OSAS; extracted the clinical symptoms, syndromes, polysomnographic findings and variables, and treatment options, and reviewed the authors' recommendations.

**Data Synthesis:** Orthodontic and craniofacial abnormalities related to pediatric OSAS are commonly ignored, despite their impact on public health. One area of controversy involves the use of a respiratory disturbance index to define various abnormalities, but apneas and hypopneas are not the only abnormalities obtained on polysomnograms, which can be diagnostic for sleep-disordered breathing. Adenotonsillectomy is often con-

sidered the treatment of choice for pediatric OSAS. However, many clinicians may not discern which patient population is most appropriate for this type of intervention; the isolated finding of small tonsils is not sufficient to rule out the need for surgery. Nasal continuous positive airway pressure can be an effective treatment option, but it entails cooperation and training of the child and the family. A valid but often overlooked alternative, orthodontic treatment, may complement adenotonsillectomy.

**Conclusions:** Many complaints and syndromes are associated with pediatric OSAS. This diagnosis should be considered in patients who report the presence of such symptoms and syndromes.

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UNDERSTANDING OBSTRUCTIVE sleep apnea syndrome (OSAS) in children requires knowledge of the physiology of sleep and breathing. There is an immediate increase in upper airway resistance with sleep onset, with an initial "overshoot" in this resistance that decreases very quickly. Still, this resistance during established sleep is mildly higher than during wakefulness.<sup>1</sup> There is also a slight decrease in tidal volume with sleep. This decrease will be more pronounced with the occurrence of rapid eye movement (REM) sleep. These mild decreases will be compensated by a slight increase in breathing frequency to keep minute ventilation normal. Breathing frequency decreases during the first 2 years of life but stays the same thereafter; it has been calculated to range from a maximum of 16 to 18 breaths/min in non-REM sleep and 17 to 19 breaths/min during REM sleep.<sup>2,3</sup>

The obesity epidemic, evident in the United States and industrialized countries, has complicated the investigation of obstructive sleep apnea (OSA) and related syndromes. Fat distribution varies according to genetic, sex, and hormonal patterns and the

inherent relationship among these 3 factors. It is common for fat to deposit in the abdominal region. Such abdominal obesity will lead to chest-bellows impairment, as seen in restrictive thoracic disorders. Although it may not lead to upper airway obstruction, abdominal obesity may worsen the poor gas exchange that may already exist because of OSAS. Sleep will always worsen the gas exchange in these subjects when they are in the supine position and when they achieve REM sleep. During REM sleep, the associated atonia eliminates contractions of the accessory respiratory muscles and the abdominal muscles, which engage in active expiration.<sup>2,3</sup> Also, REM sleep is associated with further flattening of the diaphragm's position.<sup>2</sup> These physiological changes worsen gas exchange in subjects with abdominal obesity and may even lead to REM sleep-related hypoventilation with some degree of carbon dioxide (CO<sub>2</sub>) retention. Upper airway impairment, per se, is not directly related to this CO<sub>2</sub> retention. It has, however, been hypothesized that abnormal gas exchange during sleep may impair the coordination of time-related contractions of both upper airway dilator muscles and inspiratory muscles.

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**Table 1. Complaints Reported by Parents Regarding Their Children**

Age Group and Age			
Infants, 3-12 mo	Toddlers, 1-3 y	Preschool-aged Children	School-aged Children
Disturbed nocturnal sleep with repetitive crying	Noisy breathing or snoring	Regular, heavy snoring	Regular, heavy snoring
Poorly established day/night cycle	Agitated sleep or disrupted nocturnal sleep	Mouth breathing	Agitated sleep
Noisy breathing or snoring	Crying spells or sleep terrors	Dropoling during sleep	Abnormal sleeping positions
Nocturnal sweating	Grouchy and/or aggressive daytime behavior	Agitated sleep	Insomnia
Poor suck		Nocturnal awakenings	Delayed sleep phase syndrome
Absence of normal growth pattern or failure to thrive	Daytime fatigue	Confusional arousals	Confusional arousal
Observation of apneic events	Nocturnal sweating	Sleepwalking	Sleepwalking
Report of apparent life-threatening event	Mouth breathing	Sleep terrors	Sleep talking
Presence of repetitive earaches or URI	Poor eating or failure to thrive	Nocturnal sweating	Persistence of bed-wetting
	Repetitive URI	Abnormal sleeping positions	Nocturnal sweating
	Witnessed apneic episodes	Persistence of bed-wetting	Hard to wake up in the morning
		Abnormal daytime behavior	Mouth breathing
		Aggressiveness	Dropoling
		Hyperactivity	Morning headache
		Inattention	Daytime fatigue
		Daytime fatigue	Daytime sleepiness with regular napping
		Hard to wake up in the morning	Abnormal daytime behaviors
		Morning headache	Pattern of attention-deficit/hyperactivity disorder
		Increased need for napping compared with peers	Aggressiveness
		Poor eating	Abnormal shyness, withdrawn and depressive presentation
		Growth problems	Learning difficulties
		Frequent URI	Abnormal growth patterns
			Delayed puberty
			Repetitive URI
			Dental problems appreciated by dentist
			Crossbite
			Malocclusion (class II or III)
			Small jaw with overcrowding of teeth

Abbreviations: OSAS, obstructive sleep apnea syndrome; URI, upper respiratory tract infection.

Obstructive sleep apnea syndrome was described in children in 1976.<sup>4</sup> Although children may present with OSAS, the literature had established by 1982 that children had other abnormal respiratory effort patterns during sleep that were frequently associated with snoring and clinical symptoms.<sup>5</sup>

### EPIDEMIOLOGY

No definitive population-based study has evaluated the presence of OSAS in children. Previous studies were performed in different settings and implemented a variety of tools. Some considered regular nocturnal snoring a marker of chronic obstructive breathing during sleep. The percentage of individuals younger than 18 years who have been reported with regular heavy snoring oscillated between 8% and 12%. Subjects in other studies underwent polygraphic monitoring, but these studies were limited in terms of sample size and testing difficulties; initial studies estimated OSAS prevalence to be between 1% and 3%.<sup>6-15</sup> More recently, many specialists have estimated OSAS prevalence to be between 5% and 6%. Although better monitoring techniques during polysomnography (PSG) have shown that more abnormal breathing events are present,<sup>16</sup> the definitive data are still lacking.

### CLINICAL SYMPTOMS

Abnormal narrowing in the nose, nasopharynx, oropharynx, or hypopharynx causes abnormal air exchange during sleep, which in turn leads to clinical symptoms. These symptoms will vary with age. Recognition of the problem is often only noted in older children, who are able to articulate complaints. **Table 1** indicates the parental complaints of children seen at sleep clinics over time.<sup>17-25</sup>

Abnormal breathing during sleep has been associated with specific clinical problems and findings. The clinical interview of a child suspected of having sleep-disordered breathing (SDB) must lead to systematic questioning of the parents regarding their child's symptoms; the parents may not associate the occurrence of these symptoms with abnormal breathing during sleep. **Table 2** outlines syndromes that have been shown to be related to SDB and are subsequently controlled after the appropriate treatment of the breathing disorder has been initiated.<sup>20,24,26-51</sup> Some of the syndromes are related to maxillomandibular development<sup>26</sup> and are more connected to orthodontic practice. Pediatricians do not traditionally consider orthodontic problems to be part of a child's health issues, but in light of the

**Table 2. Syndromes Related to Abnormal Breathing During Sleep**

Chronic snoring  
Daytime fatigue  
Daytime sleepiness  
Sleep maintenance insomnia  
Sleep-phase delay  
Confusional arousal  
Sleep talking  
Sleep terror  
Sleepwalking  
Enuresis (primary or secondary)  
Morning headache  
Nocturnal migraine  
Periodic limb movement  
Learning or memory problem  
Attention-deficit/hyperactivity disorder  
Abnormal social contact (psychologically withdrawn)  
Depressive affect  
Hypotension with orthostasis  
Fainting (rare)  
Hypertension (rare)  
Cor pulmonale (rare)  
Nocturnal asthma or nocturnal wheezing  
Crossbite  
Pathologic overjet  
Overcrowding of teeth  
Impacted wisdom teeth

related health care cost and syndromic association, they should.

#### CLINICAL EVALUATION AND DIAGNOSIS OF SDB

Sleep-disordered breathing in a child will be suspected on the basis of parental complaints. The presence of 1 of the syndromes listed in Table 2 should lead to a thorough interview of the behavior during sleep as well as sleep-related factors associated with SDB.<sup>17-50</sup>

The suspicion of SDB indicates the need not only for a general pediatric evaluation but also for a thorough evaluation of the upper airway anatomy. Clinically, it involves a comprehensive examination of its successive segments. Starting with the nose, one should look for asymmetry of the nares, a large septal base, collapse of the nasal valves during inspiration, a deviated septum, or enlargement of the inferior nasal turbinates (**Figure 1**). Next, the oropharynx should be examined for the position of the uvula in relation to the tongue. The scale developed by Mallampati et al<sup>52</sup> scale may help to evaluate this position. The size of the tonsils should be compared with the size of the airway; application of a standardized scale is useful.<sup>53</sup> The presence of a high and narrow hard palate, overlapping incisors, a crossbite, and an important ( $>2$  mm) overjet (the horizontal distance between the upper and lower teeth) are indicative of a small jaw and/or abnormal maxillomandibular development. This clinical evaluation provides important details of the upper airway anatomy and identifies anatomical risk factors that can predispose one to development of abnormal breath-



**Figure 1.** Illustration of many anatomical abnormalities in a 7-year-old child, including asymmetry of the nares, an enlarged septal base, large medial crus, deviation of the septum to the right, and a narrow and high-arched palate. A rapid maxillary distractor has been placed to widen the maxillary cavity, decrease the height of the soft palate, and enlarge the bony aspects of the nose.

ing during sleep.

The results of this examination must be summarized because the different anatomical narrowings have additive effects. The apparent sizes of tonsils and adenoids are not the only anatomical findings that determine whether or not SDB is present. A change in flow due to an abnormal nose, secondary development of turbulence, and the increased collapsibility at specific vulnerable points in the upper airway are elements to consider.

A complex interaction occurs between nasal breathing and maxillomandibular growth. Abnormal nose breathing in very young individuals leads to an increase in nasal resistance and mouth breathing with secondary impairment of maxillomandibular growth,<sup>54-62</sup> as shown experimentally in young rhesus monkeys.<sup>63</sup> The first 4 years of age are of particular importance because 60% of the adult face is built during that period.<sup>64</sup> Otolaryngological and orthodontic data have clearly demonstrated the impact of enlarged tonsils, adenoids, and nasal turbinates and upper airway allergies on orofacial growth in children.<sup>21,55-70</sup>

Other factors may be considered. Neck circumference and the presence of fatty infiltration should be noted,

but no scale correlates neck circumference with age or pathologic findings. The overall aspect of the face can be appreciated. The frontal aspect of the face can be subdivided into superior, middle, and inferior portions. These portions are approximately the same length in a normal child. The upper part of the bridge of the nose and the part just below the nares represent the middle third of the face. In individuals with a maxillomandibular risk factor for OSA, the lower third of the face may be longer than expected. The terms *long face* and *long-face syndrome* have been used.<sup>21,26</sup>

## OBJECTIVE CONFIRMATION OF SDB

Testing during sleep is the only way to confirm the presence of SDB. Controversy exists concerning the need for and type of test to be performed. Some of the measures used for this testing include questionnaires and scales, home monitoring, and PSG.<sup>71-74</sup>

Questionnaires with specific emphasis on the common symptoms associated with SDB have been implemented. Although questionnaires may be helpful in directing the attention of parents to the diurnal and nocturnal symptoms of SDB, the sensitivity and specificity of the questionnaires are not sufficient for affirming the presence of SDB.<sup>23,75-77</sup>

Home monitoring with or without videotaping has also been used. Ambulatory monitoring with recording of cardiac and respiratory variables has been suggested as the first diagnostic step in testing for SDB. These devices can detect the presence of drops in oxygen saturation ( $\text{SaO}_2$ ), apneas, and hypopneas; affirm the diagnosis of SDB; and lead to treatment. Associated videotaping may confirm abnormal breathing behavior. This approach may recognize severe SDB but fails to identify the presence of associated sleep disorders and partially obstructed breaths. A negative test result does not rule out the diagnosis of SDB and must be followed by PSG; however, a positive finding may lead to faster treatment.<sup>78-80</sup>

Polysomnography is the only test that may exclude the diagnosis of SDB. It must always include monitoring of sleep/wake states through electroencephalography (EEG), electro-oculography, chin and leg electromyography, electrocardiography, body position, and appropriate monitoring of breathing. A nasal cannula-pressure transducer, oral thermistor, chest and abdominal belts, a neck microphone, and pulse oximetry are recommended, but variable montages are used.

Respiratory efforts can be investigated by a variety of means during PSG. Although infrequently used, the best approach involves measuring esophageal pressure (Pes) movements. A less reliable approach is to monitor intercostal/diaphragmatic electromyography. A recently developed analysis of this signal appears promising but needs further testing in children.<sup>81</sup> Levels of  $\text{CO}_2$  may be monitored using a nasal cannula with measurement of end-tidal  $\text{CO}_2$  levels. However, the combination of 2 cannulas in the nose of a child may disturb sleep and have a negative impact on nasal breathing; thus, a transcutaneous  $\text{CO}_2$  electrode will often be needed for this measurement.<sup>16,82,83</sup>

Sleep-disordered breathing has consequences related to the repetitive changes induced by a decrease in the size

of the upper airway during sleep. As a compensatory first step, there will be an increase in breathing frequency (tachypnea) and in respiratory efforts.<sup>5,84,85</sup> The selected response is related to the decrease in size of the upper airway and the age of the subject. Following the classic equation of breathing frequency  $\times$  tidal volume = minute ventilation, tachypnea is a more common finding in young children with small and relatively unstable chests, a population with mild to moderate breathing impairment during sleep.<sup>5,84,85</sup> Despite better chest stability, this response will also be seen in older children. Tachypnea and an increase in inspiratory efforts have been seen in the same children in association with airflow limitation. The mechanisms behind a specific response and the relationship with sleep state are unknown.

The repetitive challenges resulting from a reduction of upper airway size have negative consequences on a child's well-being. However, the normative data for many of the studied variables are still unclear. The polygraphic normative data on sleep duration and sleep stages are available in children 7 years and older.<sup>86</sup> The frequency of short arousals during sleep (ie, EEG arousals lasting  $\geq 3$  seconds that can be reliably scored by 3 years of age<sup>87</sup>) is unknown for different age groups, but abnormal breathing patterns during sleep have been identified (**Table 3**).<sup>85</sup>

## INTERPRETATION OF THE PSG

There are controversies concerning PSG<sup>71,84</sup> because many existing criteria are based on information obtained from small studies. Other recommendations were taken by consensus, which means they were not necessarily based on data; still others were based on information collected with outdated technology. The specificity and sensitivity of many of the indices used have never been calculated. Only 1 study has looked at polygraphic respiratory patterns, their frequency of occurrence, their change in frequency with treatments, and their impact on the clinical outcome associated with polygraphic changes in prepubertal children.<sup>84</sup>

One of today's most debated issues is what type of respiratory event should be scored and tabulated. Another issue is determining when "pathology" is present.<sup>23,76,84</sup> Historically, the presence of OSA was easy to recognize with simple albeit relatively insensitive equipment (thermistors). Based on the variability of breathing frequency from birth to 2 years of age, an apnea was defined as "longer than 2 breaths." For many years, there was a consensus that OSA, a complete cessation of air exchange at the nose and mouth, was associated with clinical symptoms. It was shown that removal of the obstructive apnea led to improvement of the symptoms. The initial criterion for an abnormal PSG finding was at least 1 obstructive apnea per hour of sleep.<sup>89</sup>

However, pathologic findings also occurred without complete absence of air exchange. To improve the scoring system, clinicians used the term *hypopnea*, but there is no consensus as to what a hypopnea is. Following adult criteria and using thermistors with limited sensitivity, clinicians suggested that a hypopnea should last "longer than 2 breaths."<sup>4</sup> Also, the airflow signal from the combined



**Table 3. Abnormal Breathing Patterns**

Term	Definition
Apnea	Absence of airflow at nose and mouth for >2 breaths, independent of desaturation or change in EEG; subdivided into central, mixed, or obstructive apnea based on airflow and Pes recording
Hypopnea	Reduction by $\geq 50\%$ in nasal flow signal amplitude for $\geq 2$ breaths; scored independently from $\text{SaO}_2$ drop or EEG arousal; often but not always associated with snoring.
Abnormal respiratory effort	Reduction in nasal flow of $<50\%$ with flattening of nasal cannula signal (flow limitation) <sup>7</sup> and decrease in the mouth signal (thermistor); often seen with snoring and increased effort shown on Pes signal defined as Pes crescendo or continuous sustained effort
Pes crescendo <sup>a</sup>	Sequence of $\geq 4$ breaths that show increasingly negative peak end-inspiratory pressure; may be seen with flow limitation on nasal cannula
Continuous sustained effort <sup>a</sup>	Repetitive, abnormally negative peak end-inspiratory pressures, ending at same negative inspiratory pressure without a crescendo pattern; associated with discrete flow limitation on nasal cannula–pressure transducer signal, with “flattening” of the breath signal curve for $\geq 4$ successive breaths
Pes reversal <sup>b</sup>	Termination of abnormal increase in respiratory effort with abrupt switch to a less negative peak end-inspiratory pressure.
Respiratory event–related arousals	As defined by the American Academy of Sleep Medicine, patterns of progressively negative pressure terminated by a sudden change in pressure to a less negative level and an arousal event lasting $\geq 10$ s <sup>88</sup>
Tachypnea	Increase in respiratory rate, above that seen during quiet unobstructed breathing, by $\geq 3$ breaths/min in non-REM sleep, or 4 breaths/min in REM sleep, for $\geq 30$ s; no changes in $\text{SaO}_2$ , Pes, or EEG were required <sup>11</sup>

Abbreviations: EEG, electroencephalography; Pes, esophageal pressure; REM, rapid eye movement;  $\text{SaO}_2$ , oxygen saturation.

nasal-oral thermistors should decrease by at least 50% of normal baseline breathing. Hypopneas should be terminated with an EEG arousal or a drop in  $\text{SaO}_2$  of at least 3%.<sup>23,76</sup> By these criteria, the finding was considered to be pathologic if the obstructive apnea index was at least 1 or if the apnea-hypopnea index was at least 5 events per hour.

Some children with very noisy breathing at night and enlarged tonsils and adenoids had a normal score at PSG but had clinical symptoms<sup>90,91</sup> that led to adenotonsillectomy. Also, other SDB syndromes without an associated abnormal apnea-hypopnea index but with an elevated respiratory disturbance index (RDI) were controlled with nasal continuous positive airway pressure (CPAP) or upper airway surgery.<sup>85</sup> Although an apnea-hypopnea index of at least 5 was considered pathologic, there was the recognition that apnea and hypopnea as defined did not encompass all pathologic breathing during sleep. Hence, an arousal index was calculated; thus, snoring sequences that were terminated with an EEG arousal were scored. The association of apnea-hypopnea and other measurements led to the use of the term *RDI*. This term acknowledges that the defined PSG patterns did not encompass all abnormal breathing events.

The introduction of the nasal cannula–pressure transducer system<sup>16,92</sup> allowed a more accurate recognition of abnormal breathing during sleep, as this technique based on nasal flow is semiquantitative. It allows better recognition of partially obstructed breaths. A hypopnea was defined when flow decreased by 30% of a normal breath. However, many still require an EEG arousal and/or an  $\text{SaO}_2$  drop, despite previous demonstration that clinical consequences can be obtained without a change in  $\text{SaO}_2$ . An RDI of more than 5 events was used on the basis of previous habits.

A minority of sleep clinics monitored respiratory efforts using Pes. These groups showed that snoring without hypopneas was associated with abnormal efforts and an induction of EEG arousals. Based on Pes record-

ings,<sup>83,85</sup> specific patterns were recognized and defined, such as “Pes crescendos,” “sustained respiratory effort,” and “Pes reversals.” Some evidence suggests that these patterns were frequently, but not always, seen with abnormal nasal flow on the nasal cannula–pressure transducer recording. However, a flow limitation ranging from normal to a 30% decrease at the nasal cannula was usually seen with these patterns. The nasal flow limitation was described as a flattening of the nasal flow curve; several patterns of abnormal curves have been described. It may be easier to visually recognize a change of the Pes than a flattening of the nasal curve.<sup>83,85</sup>

The application of these Pes-related definitions showed that children who had no apneas or hypopneas,  $\text{SaO}_2$  drop of 3% or more, or EEG arousals presented with clinical complaints and clinical sleep-related syndromes, primarily parasomnias.<sup>32,33</sup> By applying these criteria, a clinical outcome study performed at the Stanford University Sleep Disorders Clinic, Stanford, Calif, focused on clinical complaints and the presence of clinical symptoms and signs. Complete treatment of the sleep-related upper airway problem with resolution of symptoms and signs was associated with fewer than 1 of the events included in the RDI.<sup>84</sup> Persistence of symptoms and signs was associated with the continued presence of an event that was not necessarily an apnea or a hypopnea. Instead, the breathing event was noted to be a “flow limitation with an increase in respiratory effort” or merely an increase in respiratory effort, and a cutoff point for the RDI of greater than or equal to 1.5 events per hour of sleep was found.<sup>84</sup> However, an RDI of greater than or equal to 1.5 events per hour is based on only 1 outcome study, even if several clinical studies have indicated the validity of such a cutoff point.<sup>32,45,84</sup>

## ANS AND BREATHING PATTERNS DURING SLEEP

An increase in respiratory efforts is associated with changes in autonomic nervous system (ANS) settings. These changes will affect the cardiovascular system in an indi-

vidual with a normal ANS.<sup>49</sup> One may want to evaluate these changes to recognize an abnormal pattern and determine whether they may be detrimental. The following 2 types of responses can be seen when an increase in respiratory effort occurs during sleep: activation or arousal with cortical involvement.

*Activation* is a clinical neurophysiology term defined by Moruzzi and Magoun<sup>93</sup> during the course of their study of the ascending reticular formation; it is related to the recruitment of sensory inputs that will lead to a polysynaptic motor response after relay of sensory input in the brainstem and subcortical structures. The nucleus ambiguus receives information that simultaneously leads to efferent responses through the nucleus tractus solitarius. This relay leads to a simultaneous ANS stimulation, and an autonomic activation will lead to an increase in sympathetic tone.

An ANS response may be seen with brainstem reflexes leading to full reopening of the upper airway without EEG cortical arousal, or it may be seen as the consequence of an EEG cortical arousal. The presence of cortical arousals will be associated with clinical symptoms, such as complaints of excessive daytime somnolence, irritability, or unrefreshing sleep. The role of repetitive activation is unknown in children.

The determination of how much airway size changes and the duration of the change needed to lead to cortical arousal are unknown. Sleep stages may play a role in the type of response seen, but no definitive information is available in prepubertal children.

The pulse transit time, which measures the transit time of the pulse wave from approximately the aortic valve to the wrist, and the peripheral arterial tonometry are 2 variables that were added to PSG to help recognize an arousal.<sup>94-97</sup> None of these devices can distinguish between a brainstem reflex and a cortical arousal response. The importance of the sympathetic response could be a relatively accurate indicator of cortical involvement, but the studies to validate such distinction have not yet been published. Based on a commercially designed algorithm involving heart rate and finger plethysmography, the peripheral arterial tonometry does not really reflect the balance between the sympathetic and parasympathetic systems during sleep. The pulse transit time also has limitations of interpretation. When used to identify cortical arousals related to SDB, both techniques have false-positive and false-negative findings, which limit the accuracy of interpretation.<sup>97</sup> The monitoring of these different variables has, however, shown that repetitive snoring can be associated with activation and/or EEG arousal.

## CHANGES IN EEG SLEEP PATTERNS

Historically, an EEG alpha or an alpha and beta arousal lasting 3 seconds at the termination of an abnormal breathing event was required to score an event. However, several studies have shown that limited upper airway occlusion may end with a burst of delta waves or a K complex.<sup>98</sup> The use of a sleep scoring system, based on analysis of the cyclic alternating pattern (CAP), demonstrated the negative effect of these bursts.<sup>99-101</sup> The CAP scoring system is based on recurrent bursts of delta waves and K com-

plexes with or without superimposed alpha waves within a period of 60 seconds intertwined with low EEG amplitude. The CAP is a normal phenomenon that occurs between wakefulness and slow-wave sleep or, at the end of night, between REM sleep and well-established, repetitive sleep-spindle sleep. It indicates a transition from one stable state to another and is not seen in REM sleep. CAP is typically a transient period during which a greater instability of sleep may occur with a greater chance to enter a light sleep or even to awaken. An abnormal CAP rate, defined in different age groups in children,<sup>100,101</sup> indicates an instability of non-REM sleep as well as a difficulty in reaching a new stable state.<sup>99</sup> CAP is associated with autonomic activation and may lead to awakening and large sympathetic discharge. Chervin et al<sup>102,103</sup> have also reported a novel approach to evaluate EEG findings with abnormal breathing during sleep, based on an algorithm investigating the EEG changes seen, with each abnormal inspiration associated with increased effort. The algorithm recognizes the changes in brain wave activity with increased inspiratory effort. When adenotonsillectomy is successful in relieving abnormal breathing during sleep, the abnormal EEG pattern disappears. Furthermore, the daytime symptoms, particularly sleepiness, abate. This analytic technique needs to be tested further.

## GENETIC RISK FACTORS OF SDB

Genetic<sup>64,104-112</sup> and environmental risk factors have been identified in the development of SDB; they are associated to variable degrees. Oral mucosa thickness has been identified as an ethnic risk factor in African Americans, and skull base length has been noted to be an ethnic risk factor in Far East Asians.<sup>108,109</sup> African American and Far East Asian populations have been shown to have significantly higher risk than Caucasians when age, sex, and body mass index are considered.<sup>64,104-112</sup> The familial trait of dolichocephaly (or narrow face) has also been implicated as a risk factor, independent of ethnicity.<sup>51,111</sup> Familial cases of SDB are seen in all ethnic groups. Genetic investigations are performed, although there is currently no clear indication for a specific gene location responsible for increased risk. The strongest current indicators have been related to facial morphotype.<sup>64</sup> Clearly, there is an increased risk of SDB in a family in which a member is affected.<sup>104-106,110-112</sup> Pediatricians should, therefore, systematically question other family members about sleep-related problems when there is a positive history of SDB (**Figure 2**).

## TREATMENT

There is an overall consensus that children with SDB should undergo evaluation by an otolaryngologist for surgical treatment. It is also clear that the well-described but extremely complex interaction between nasal breathing and facial growth is important, even if it is rarely investigated.<sup>51</sup>

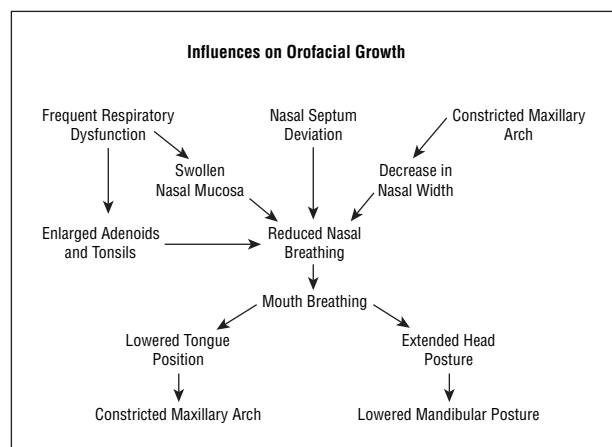
Treatment for short-term outcomes indicates that adenotonsillectomy with or without radio frequency treatment of nasal inferior turbinates is the first approach to consider.<sup>85,113,114</sup> Independent of the size of tonsils or adenoids, adenotonsillectomy will provide more airway

space. Two points must be emphasized. First, outcome investigation has shown that isolated tonsillectomy or adenoidectomy is not as effective as adenotonsillectomy.<sup>84,115</sup> Also, if enlarged turbinates are present, radio frequency treatment of the nasal turbinates should be performed at the same time, while the child is under general anesthesia. Performance of adenotonsillectomy without performance of nasal turbinate treatment may have a negative impact on the outcome.<sup>84</sup> Outcomes of adenotonsillectomy have been reviewed,<sup>113</sup> but no review addresses the reasons for failure. A recent study examined the short-term outcomes to understand why results were incomplete.<sup>115</sup> Surgeons often use techniques that are not aimed at maximally opening the airway; they may fail to treat the nose simultaneously with the adenotonsillectomy; and others simply do not recognize the craniofacial changes that contributed to the SDB.

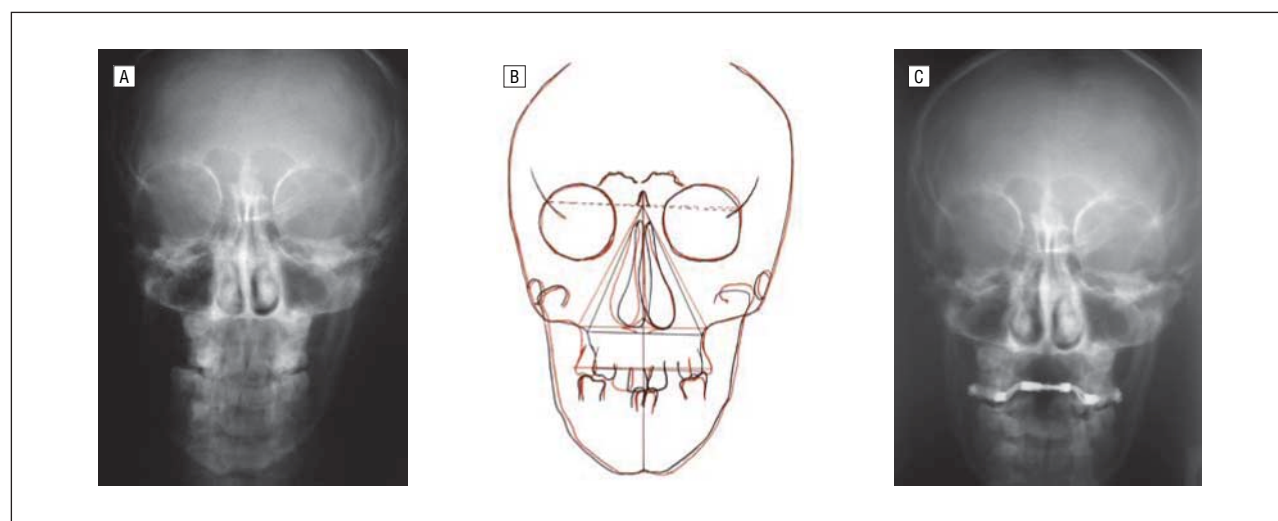
Only 2 studies<sup>116,117</sup> have looked at the long-term outcome of regular adenotonsillectomy performed in prepubertal children. After evaluating the outcomes to a minimum of 10 years later, both studies indicated that there was failure to control the problem because of the pres-

ence of hypopneas and apneas at the long-term follow-up recordings. Demonstration of the absence of apneas and hypopneas within 6 weeks to 3 months after surgery was requested in 1 of the 2 studies.<sup>116</sup> The long-term outcome in that study linked the recurrence of abnormal breathing during sleep to the absence of dealing with a narrow maxilla and/or mandible at the time of the initial surgery and the later occurrence of tongue/mucosal enlargement at the time of puberty, when 90% of orofacial adult growth had already occurred.

Adenotonsillectomy has been performed in association with orthodontic treatment.<sup>118</sup> Rapid maxillary distraction (RMD) is an orthodontic technique that is based on the bone formation process. A distractor anchored to 2 molars on both sides applies daily pressure, pushing apart both halves of the maxilla; bone then grows from the borders of the cartilage.<sup>118,119</sup> This technique pushes the soft tissues laterally, decreases the height of the soft palate, and enlarges the nasal orifices.<sup>118</sup> Rapid maxillary distraction may be associated with distraction of the mandible, but because no mid-cartilage is present, there is very limited widening. This fact may limit the degree of maxillary widening with RMD (**Figures 1, 3, and 4**). Slow maxillary distraction is based on similar principles and optimizes the degree of widening at the different growth periods that occur in prepubertal children. Rapid and slow maxillary distractions are performed between 5 and 11 years of age. Distraction results in widening of the palate and the nose; thus, this procedure remedies nasal occlusion related to a deviated septum, for which little can be done before 14 to 16 years of age. Even when performed in association with adenotonsillectomy, however, orthodontics may not control all SDB. Abnormal mandibular or maxillomandibular anteroposterior development is a bigger challenge. Nasal CPAP will be the recommended treatment until further orthognathic surgery<sup>120</sup> can be performed.

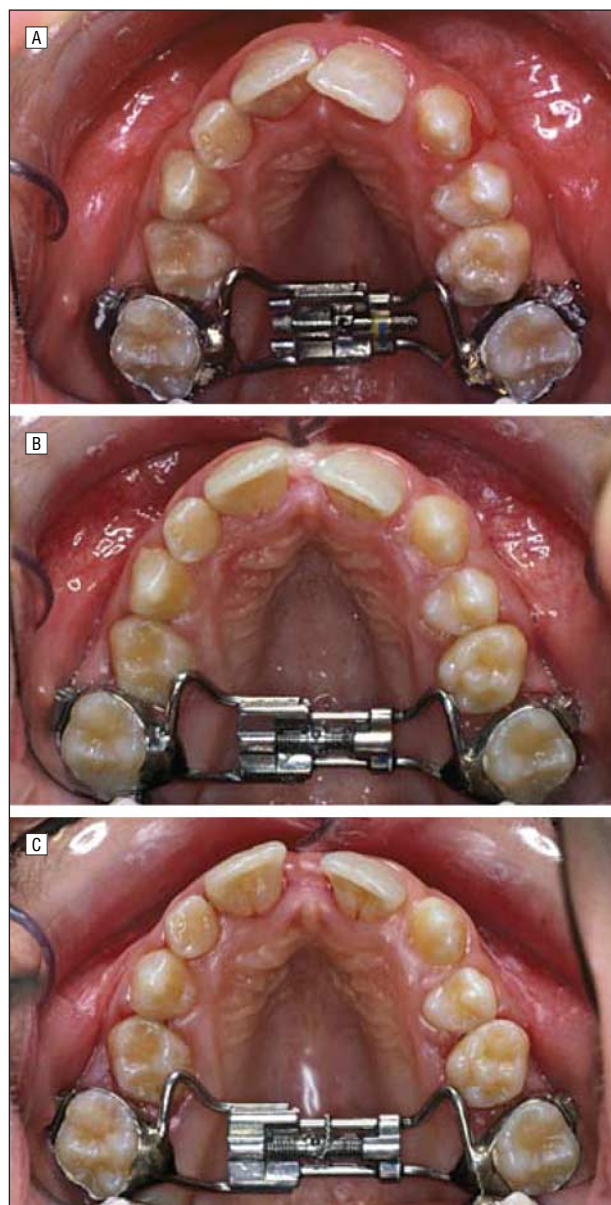


**Figure 2.** Influences of sleep-disordered breathing on orofacial growth.



**Figure 3.** The significant impact of rapid maxillary distraction. A, Frontal cephalometric study demonstrates a narrow maxillary arch before distraction. B, Drawing superimposing the image in A and the postdistraction image (shown in C) to show the widening of the maxillary and nasal cavities. The patient's right inferior turbinate is closely approximated to the septum. C, The frontal cephalometric demonstrates that the maxillary arch has been opened since the distractor has been placed. The nasal cavity has also been altered because the patient's right inferior turbinate is now farther from the septum than it was before placement of the distractor.





**Figure 4.** Rapid maxillary distraction demonstrates progressive improvement in the crowding of a child's teeth, from immediately after insertion of the distractor (A) to 3 weeks later (C). Progressive widening is indicated by the space between the 2 frontal incisors.

Home nasal CPAP has been used in infants, prepubertal children, and pubertal children. The first report of its usefulness in children in 1986 was a prospective study that followed up 5 children, aged 3 to 11 years, for 10 months.<sup>121</sup> Similar findings were reported in several large retrospective studies (**Table 4**).<sup>103,122-127</sup> These studies primarily involved children older than 12 months. Infants aged 8 to 18 weeks were followed up from the onset of treatment through the first 12 months of age in a study in 1995<sup>124</sup>; that study was replicated in 1999.<sup>126</sup>

The difficulty in the application of nasal CPAP relates to training the family and child and finding the appropriate nasal interface. Children often need to be trained to tolerate the facial interface. (Behavioral modification techniques and daytime training may help with this training.) Continuous positive airway pressure is very useful when the SDB is related to major craniofacial deformities or other illnesses. If the upper airway problem is complicated by neuromuscular disease, nasal bilevel treatment may be used.

Regular follow-up should be performed within the first 3 months to evaluate mask fitting.<sup>124</sup> Because of rapid craniofacial growth of young children, CPAP should be evaluated every 6 months. An annual visit with a craniofacial specialist should occur to affirm that the headgear and mask do not worsen a maxillary growth deficiency.<sup>128</sup> Clinicians should encourage the use of humidification, aggressively treat allergies and/or rhinitis, and check nasal patency. In light of children's favorable response to surgery with or without orthodontic and antiallergic treatment, nasal CPAP should only be a second consideration.

Orthognathic surgery entails shifting bones and disrupting the bone growth structures.<sup>120</sup> Such an approach is normally postponed until 10 to 13 years of age. Two surgical techniques used in patients with SDB are mandibular distraction osteogenesis and maxillomandibular advancement.

**Table 4. Nasal CPAP in Children**

Source	Type of Study	PSG	Conclusions
Guilleminault et al, <sup>121</sup> 1986	Feasibility study of 5 children in hospital; prospective 10-mo home study of 5 children	Before, during titration, and during follow-up	Feasibility with parent training; 4 of 5 infants daily use of CPAP at 10-mo; follow-up
Waters et al, <sup>122</sup> 1995	Retrospective review of 80 children aged 12 d to 15½ y	For diagnosis and titration	86% of parents completed training; 12.5% dropped out
Marcus et al, <sup>123</sup> 1995	Retrospective study of 94 children aged 3-12 mo; applied after adenotonsillectomy in 76%; first treatment in 23 children	For diagnosis and titration	1 Dropout
Guilleminault et al, <sup>124</sup> 1995	Prospective study of infants aged 8-18 wk at entry and systematic follow-up for 12 mo; family underwent screening at entry for understanding of treatment	For diagnosis, treatment, and each follow-up retitration	Need to readjust equipment and pressure on regular basis owing to fast craniofacial growth in infancy
Rains, <sup>125</sup> 1995	Prospective study of 4 children aged 3-12 y; training of parents	For diagnosis and titration	Follow-up for 3 mo; effective treatment; no dropout for 3 mo; 1 dropout thereafter
McNamara and Sullivan, <sup>126</sup> 1999	Prospective study of 24 infants aged 6-51 wk for 12 mo	For diagnosis, titration, and regular follow-up	Family training and support; continuous use in 18 children; effective treatment
Downey et al, <sup>127</sup> 2000	Retrospective study of 18 children aged <2 y		12 Children successfully treated

Abbreviations: CPAP, continuous positive airway pressure; PSG, polysomnography.



Mandibular distraction osteogenesis is very similar to RMD, but it is applied to the mandible when a maxillary and mandibular widening are needed and when the slow mandibular orthodontic distraction cannot achieve the needed result.<sup>120</sup> A vertical transection of the maxilla is performed between the 2 central incisors and a distractor is used as in RMD. Widening of 12 to 14 mm can be obtained easily in 3 weeks. Orthodontic treatment is similar to that described with RMD. By 12 to 13 years of age, both procedures can be performed simultaneously to provide an anterior displacement of the tongue and enlargement of the retrolingual airway space.<sup>120</sup>

Maxillomandibular advancement is a very successful procedure. Nevertheless, it is major surgery that should be performed after there has been appropriate orthodontic treatment. Surgeons who perform this procedure must have a good understanding of upper airway mechanics and dental problems. It may be performed at any time during childhood, but it is often postponed until 11 to 12 years of age.<sup>129</sup>

A controversial issue is how early to perform adenotonsillectomy. Most will agree that adenotonsillectomy is often performed by 24 months of age. However, OSA has been noted as early as 3 weeks of age, and cases of heavy snoring and clinical symptoms in children aged 6 to 24 months are actually common. Adenotonsillectomy has been performed as early as 6 months of age.<sup>112</sup>

Several advances have been made in sleep medicine. Apneas and hypopneas are not the only indicators of abnormal breathing during sleep. In this rapidly evolving field, it has been challenging to establish new scoring criteria, despite the availability of new technologies. However, the clinical findings and the PSG results should be used to determine the diagnosis and to guide treatment recommendations.

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