Obstructive Sleep Apnea in Children: Controversies in Diagnosis and Management

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ABSTRACT

Aim: To discuss commonly encountered diagnostic and therapeutic dilemmas in pediatric obstructive sleep apnea (OSA).

Background: Pediatric OSA is a fairly common childhood disorder, affecting 1 to 5% of all children and much larger proportions of children with selected comorbidities. Untreated OSA is associated with deleterious effects on neurobehavioral outcomes, cardiovascular health, and growth.

Results: We discuss several important diagnostic dilemmas, including when to obtain a preoperative polysomnogram (PSG), the relationship between OSA and sleep-disordered breathing, and limitations of conventional PSG-derived metrics. Management challenges commonly encountered in clinical practice include defining surgical cure and providing reliable estimates for families preoperatively, issues related to the use of positive airway pressure, and emerging alternative and complementary therapeutic modalities.

Conclusion: While recently published clinical practice guidelines have provided important standards for the diagnosis and management of pediatric OSA, many areas of uncertainty remain.

Clinical significance: We provide a review of current diagnostic and therapeutic controversies relevant to the practicing clinician.

Keywords: Obstructive sleep apnea, Pediatric, Polysomnography.

INTRODUCTION

Obstructive sleep apnea (OSA) is a relatively common pediatric disorder and is associated with adverse health consequences including neurocognitive and behavioral problems, cardiovascular complications including systemic and pulmonary hypertension, and impaired growth. Although recent clinical practice guidelines have been published by both the American Academy of Pediatrics (AAP) and the American Academy of Otolaryngology—Head and Neck Surgery (AAO-HNS) guidelines, an evidence gap still exists to direct appropriate diagnosis and treatment. In this article we discuss selected commonly encountered controversies in the diagnosis and management of pediatric OSA.

DIAGNOSTIC DILEMMAS

To make the Diagnosis of OSA, does One require a Sleep Study? Yes

The International Classification of Sleep Disorders—3rd edition states that one must have an abnormal sleep study in combination with either nighttime or daytime symptoms. Nighttime symptoms may include habitual snoring, witnessed apneas, mouth breathing, sweating during sleep, restless sleep, nocturnal enuresis (secondary), sleepwalking, night terrors, and neck hyperextension. Daytime symptoms include daytime sleepiness, hyperactivity, poor school performance, inattention or impaired concentration, aggressive behavior, difficulty waking in the morning, and headaches that are worse in the morning.

Can a Child meet the AASM Definition of OSA but have an OAHI < 1 Event/Hour? Yes

Partial chronic upper airway obstruction exists when there are significant gas exchange abnormalities without discrete respiratory events. Pediatric OSA may be diagnosed when there is a supportive clinical history in conjunction with the following polysomnogram (PSG) findings: (a) obstructive apnea-hypopnea index...
Does Tonsil Size matter? Yes and No

One is more confident that a child may have SDB secondary to the tonsils when they are hypertrophic (3+ to 4+), although previous studies have demonstrated a weak relationship between tonsil size and OSA severity.2 The questionable importance of tonsil size is further highlighted in the Childhood Adenotonsillectomy Trial (CHAT) study since the qualification for study entry required the presence of a visible tonsil (1+) not one that was hypertrophied.

Can a Child have a “Normal Sleep Study” but still benefit from Intervention?

The clinical significance of primary snoring vs OSA is one issue at the core of the controversy related to the appropriate role of preoperative PSG in the otherwise healthy child with clinical SDB and tonsillar hypertrophy. Preoperative PSG is able to differentiate children who are primary snorers from those with OSA. Whether or not this distinction is clinically important depends on differences in associated morbidity and response to therapy. Some previous studies have demonstrated neurobehavioral deficits in children with primary snoring, and there is no clear dose response between degree of OSA and neurocognitive impairment.3 In addition, children with primary snoring have been found to clinically benefit from tonsillectomy and adenoidectomy (T&A) compared with observation.4 These previous findings are bolstered by recent results from the CHAT study, in which there was a mild, if any, relationship between OSA severity as assessed by PSG and baseline morbidity or response to T&A.5,6 Currently, the Pediatric Adenotonsillectomy Trial for Snoring (PATS) is recruiting participants with primary snoring to be randomized to either T&A or watchful waiting, and will provide more outcomes data to inform this discussion.

The observed disconnect between the OAHI and important OSA-associated clinical sequelae may represent the OAHI’s inability to fully capture work of breathing, perhaps the core pathologic feature of the disease. This view is supported by studies that utilize quantitative esophageal pressure measurement, considered the gold standard for work of breathing. Chervin et al7 demonstrated that, among children with OSA, quantitative esophageal pressure measures, not OAHI, were associated with disruptive behavior and sleepiness at baseline and their improvement after adenotonsillectomy.

Are the Normative Values reported in the ICSD-3 Valid? Probably not

There is considerable debate regarding what constitutes a normal OAHI. Previous studies that reported normal values for OAHI in children without known sleep disorders are reported in Table 1. The data are reported as the mean and standard deviation, and an upper limit of normal (ULN) calculated as the mean plus two standard deviations. These reported normal values would tend to support the cutoff of an OAHI of one per hour as abnormal. However, there are several crucial factors to consider when interpreting these previous studies, mainly related to the definition of hypopnea that was utilized (Table 1). The combination of the lack of nasal pressure transducer, 50% reduction in flow compared with the current 30% suggestion for hypopnea, and lack of standardization for the hypopnea definition leaves open the possibility that previous studies may have underestimated what constitutes a normal OAHI compared with the current modalities in use in sleep labs across the country. If one could repeat the normative studies from a decade ago using today’s scoring criteria and equipment, the threshold for normal would likely be higher.

Does the OAHI tell the Entire Story? No

Possible metrics that can be factored into severity assessment include measures of respiratory event frequency (OAHI, AI, Respiratory Disturbance Index), oxygen distribution (oxygen nadir, percent sleep time spent below 90%), and gas exchange (peak CO₂, percent sleep time spent with CO₂ above 50 mm Hg). For suboptimal studies where the child does not tolerate the flow sensors, one may use the asleap video characteristics to help determine the severity of sleep disruption. Multiple studies have demonstrated little to no relationship between OAHI and baseline neurobehavioral morbidity or response to therapy among children with OSA, with CHAT being the most recent example.5
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When is a Child no Longer a Child (i.e., when should One use Adult Scoring Criteria)?

Although the pediatric criteria for OSA apply to all patients younger than 18 years of age, respiratory events may be scored using either adult or pediatric criteria during the teenage years. The most recent American Academy of Sleep Medicine (AASM) Manual for the Scoring of Sleep and Associated Events states that criteria for respiratory events during sleep for infants and children can be used for children younger than 18 years, but it is up to the individual sleep specialist to choose if adult or pediatric rules for scoring respiratory events should be applied in those between age 13 and 17 years. The main difference in scoring rules between adults and children for respiratory events relates to the duration required for an event to be scored. Because children have faster normal respiratory rates compared with adults, obstructive apnea and hypopnea definitions are based on changes occurring over the duration of 2 breaths; in contrast, adult scoring respiratory event rules are based on events that occur over 10 seconds. Another guide point would be if the “child” has finished their maturational changes (i.e., Tanner 5).

Are there Alternative Modalities other than a PSG that have a Role in the Decision to recommend Intervention vs Observation?

Given that the OAHI and other conventional metrics obtained during PSG have limited association with OSA-associated morbidity and response to treatment, there are alternative measures currently under investigation. Experimental measures of sleep disturbance and respiratory effort include cyclic alternating patterns, duty cycle, pulse transit time, and biomarkers. While there have been some promising initial results suggesting that several of these markers may better characterize OSA and capture its associated morbidity compared with conventional metrics, ongoing research and future studies are attempting to more fully elucidate test characteristics prior to inclusion in mainstream clinical practice.

There are also several alternative devices under investigation for the diagnosis of OSA in children. Overnight pulse oximetry has been used for years, and a classification system has been developed according to the McGill Score. While an abnormal nocturnal oximetry study does provide good positive predictive value in the appropriate clinical context, the reliance on only a single channel results in low negative predictive value and cannot delineate respiratory events, in contrast to standard PSG. Home sleep studies with many or all of the same sensors included as in-lab PSG are also being investigated. Some studies have demonstrated adequate technical feasibility in children, although this is currently in the investigational phase. Finally, a recent device, named the Sonomat, has been under investigation. This is a contactless monitoring system that records body movements, breathing movements, and breath and heart sounds from a series of vibration and sound sensors embedded in a mat that the patient sleeps on in their home setting; an initial validation study in adults has yielded encouraging results.

What is the Role of Preoperative PSG?

The appropriate role of preoperative PSG remains controversial. Currently, both the AAP and the AASM advocate for routine preoperative PSG in the child with suspected OSA in order to confirm the diagnosis prior to surgery. This recommendation is based primarily on the knowledge that history and physical examination alone do not consistently predict the presence or severity of OSA based on conventional PSG metrics. In contrast, the AAO-HNS advocates for the use of preoperative PSG in children with significant comorbidities or those with discrepancy between tonsil size on examination and reported history, and recommends PSG in children at higher risk of perioperative complications. In our opinion, it makes sense to perform preoperative PSG when the child is at a higher perioperative risk, when T&A is unlikely to be curative, when parents request objective diagnosis prior to surgery, or when history and examination are inconsistent.
One argument in favor of preoperative PSG is that it can differentiate mild from moderate/severe OSA. This distinction may be clinically useful because children with mild OSA may be candidates for medical therapy with intranasal steroids and/or leukotriene inhibitor rather than surgery. There is evidence from randomized clinical trials demonstrating that children with nonsevere OSA benefit from medical therapy.\textsuperscript{13-16} Therefore, preoperative PSG may identify children with nonsevere OSA who are candidates for medical, rather than surgical, therapy.

**CHALLENGES IN MANAGEMENT**

**Surgical Cure**

Adenotonsillectomy is considered first-line therapy for OSA in children. When discussing treatment options in the clinical setting, families naturally are interested in cure rates of T&A vs other treatment options. Unfortunately, the answer depends on how cure is defined and on patient characteristics. Results from CHAT suggest normalization of PSG (defined by OAHI < 2/hour) in 79\% of children at 7 months after T&A; children with severe OSA or major comorbidities were not included in this study.\textsuperscript{17} A meta-analysis of over a thousand children demonstrated normalization of PSG (defined by AHI < 1) in 74\% of uncomplicated patients\textsuperscript{18}; in contrast, the same meta-analysis demonstrated that children with selected characteristics (severe obesity, severe baseline OSA, younger) had substantially lower rates of normalization on PSG, at 38\%. A multicenter trial demonstrated a similar pattern, with normalization of PSG significantly affected by baseline factors including age, obesity, comorbid respiratory disease, and severity of baseline OSA.\textsuperscript{19} Children with Down syndrome and craniofacial anomalies are also at increased risk for persistent OSA following T&A.

Cure rates also vary depending on the outcome of interest. Various definitions of PSG normalization have been used in previous studies (e.g., OAHI < 1, < 2, < 5/ hour). Furthermore, cure should not just be limited to PSG results, but also clinical symptoms. This was best illustrated by the watchful waiting arm of the CHAT trial, in which 42\% of children had spontaneous normalization of their PSG, but only 15\% of children had symptomatic resolution.\textsuperscript{20} Given the nuances delineated above, an accurate and understandable answer for parents inquiring regarding cure rates would be that T&A is curative in the majority of healthy children with nonsevere disease; in contrast, while children with obesity, severe baseline disease, or other selected comorbidities usually have a meaningful reduction in the severity of OSA, surgical cure is typically not achieved with T&A alone.

**Positive Airway Pressure**

While T&A is generally effective, there are several additional established and emerging treatment options for OSA in children. Positive airway pressure (PAP) is typically employed when children with moderate to severe OSA are not good surgical candidates, if the family prefers a nonsurgical option, or for residual disease following T&A. If tolerated by the child, PAP is generally very effective at relieving upper airway obstruction and treating OSA.\textsuperscript{21} There has never been a sham-controlled randomized trial for PAP in children. A single randomized trial comparing PAP with T&A was performed in a sample of children with either Down syndrome or mucopolysaccharidosis; results demonstrated substantial improvement in both groups from baseline, but no significant between-group differences at 12 months follow-up with respect to AHI, Epworth Sleepiness Scale, or OSA-18.\textsuperscript{22} Although PAP devices are only approved for those children who weigh at least 30 kg, they are clinically used in children below that weight if tolerated. While PAP is generally safe with few complications, inappropriate mask fit may result in skin irritation. In addition, long-term use may alter midface development and lead to hypoplasia. Efficacy is limited by child adherence, which may be a significant barrier, especially in children with neurodevelopmental or sensory disorders. In such circumstances, desensitization performed by a behavioral professional can be helpful. For children with respiratory compromise following a T&A, PAP in the immediate postoperative period is a management option, although there may be theoretical risk of subcutaneous emphysema.\textsuperscript{23} Finally, although auto-titrating PAP (APAP) is commonly used in adults with OSA, it is not standard practice in children. Preliminary evidence suggests that APAP does not provide optimal continuous positive airway pressure (CPAP) pressure compared with in-lab determined CPAP in children\textsuperscript{24}; we speculate that this is related to the APAP algorithms being based on respiratory events in adults.

**Alternate Therapies**

Watchful waiting is an option in children with nonsevere OSA is and mild clinical sequelae. Results from CHAT are informative regarding this approach, as 42\% of the watchful waiting arm experienced normalization of PSG but only a small minority had symptom resolution. In clinical practice, by the time children are referred to sleep or otolaryngology providers for OSA management, they are often experiencing bothersome symptoms. In ambiguous cases, it may be helpful to utilize a standardized instrument such as the OSA-18 or the Sleep-Related Breathing Disorder subscale of the Pediatric Sleep Questionnaire to assess for OSA-related symptoms and quality of life.
Several therapies may also be employed as alternatives or in addition to traditional approaches. As discussed above, intranasal steroids and/or leukotriene inhibitor may be considered in nonsevere cases. Orthodontic treatment with rapid maxillary expansion can be beneficial in children with high arched palate by widening the palate and optimizing the nasal airway; importantly, this should be performed prior to puberty when the sutures fuse. Excessive weight may contribute to OSA severity, and weight loss in obese or overweight children may lessen disease severity. If a child’s OSA is substantially worse in the supine position, positional therapy may be considered; several devices are commercially available. Myofascial reeducation, which consists of exercises that strengthen the tongue and facial muscles, has been shown to decrease residual OSA following T&A. While all of these approaches show promise, there is currently no standardized approach to their implementation. A practical algorithm which includes some of these approaches in conjunction with conventional therapies has been proposed by Kaditis et al based on PSG findings, clinical sequelae, and patient characteristics.

CONCLUSION

There have been major advancements in the diagnosis and management of OSA in children over the last decade. Clear milestones include the AASM providing standardized scoring and diagnostic criteria for PSG diagnosis of OSA specific to children, the first randomized controlled trial of T&A for OSA in children, and the development of effective treatments beyond T&A. The field will benefit from continued efforts to develop improved metrics and devices for OSA diagnosis, define appropriate treatment of primary snoring, and delineate optimal therapeutic pathways for special populations.

REFERENCES


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