



# New Options in Pediatric Obstructive Sleep Apnea

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Published online: 3 May 2019

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## Abstract

**Purpose of Review** To describe the current literature pertaining to the evaluation and management of obstructive sleep apnea (OSA) in children.

**Recent Findings** Newer diagnostic modalities, such as drug-induced sleep endoscopy (DISE) and cine MRI, may be utilized to determine anatomic locations of upper airway obstruction contributing to OSA. Pharmacologic and non-pharmacologic treatments have been studied as primary therapy for OSA or as adjuvant therapy for residual disease after adenotonsillectomy. Surgical therapy for persistent moderate to severe OSA following adenotonsillectomy may be considered for patients who are intolerant of or noncompliant with continuous positive airway pressure (CPAP) therapy and tailored to pertinent anatomic sites of upper airway obstruction.

**Summary** The current body of literature emphasizes evaluation and management for children with recurrent or persistent OSA after adenotonsillectomy. Further studies are needed to determine the long-term effectiveness of various pharmacologic, non-pharmacologic, and surgical therapies.

**Keywords** Obstructive sleep apnea · Pediatric · Management · Evaluation

## Introduction

Obstructive sleep apnea syndrome (OSAS) affects 2–4% of children, and if uncorrected, can significantly affect the quality of life and lead to neurocognitive, behavioral, and cardiopulmonary sequelae. While the majority of cases are due to adenotonsillar hypertrophy, 30–40% of children will have persistent disease after adenotonsillectomy, particularly those with craniofacial abnormalities, Down syndrome, or other comorbidities [1, 2]. Thus, the evaluation of pediatric OSAS should begin with a thorough history and physical exam with a focus on symptoms and possible causes of obstruction, including an assessment of overall body habitus, nasal anatomy, oral cavity, dentition, tongue size

and position, and tonsil size. While the overall physical characteristics and symptom profile may allude to the presence of an obstructive sleep disorder, these are poor predictors of disease presence and severity [3]. While the gold standard for the diagnosis of OSAS remains an overnight, attended, in-laboratory polysomnogram (PSG), there is considerable interest in finding alternative diagnostic modalities due to the expense, limited availability, and long wait times associated with PSG. Once diagnosed, assessment is then focused on the identification of the source of obstruction in order to guide appropriate therapy. This review will focus on newer diagnostic modalities including nocturnal pulse oximetry, peripheral arterial tonometry (PAT), drug-induced sleep endoscopy (DISE), and cine MRI, as well as

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This article is part of the Topical Collection on *Sleep Apnea*

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discuss the medical and surgical treatment options beyond adenotonsillectomy and continuous positive airway pressure (CPAP).

## Evaluation of Pediatric Obstructive Sleep Apnea

### Nocturnal Pulse Oximetry

Overnight pulse oximetry is one of the more commonly used diagnostic modalities, especially in areas with limited resources. These tests are widely available, relatively low cost, and easy to analyze with formal scoring carried out by assessing clusters of desaturation events (McGill oximetry scoring system) or the oxygen desaturation index (ODI) [4]. The McGill scoring system is a validated classification system, which takes into account the depth, number, and clustering of desaturation events. It is scored by whole numbers from 1 through 4, with correlations to AHIs of 4.1, 12.6, 13.3, and 39.9 events/hour [5]. A recent systematic review found that when the McGill scoring was applied, oximetry studies had a 97% positive predictive value (PPV) to detect an apnea-hypopnea index (AHI) > 1, but only a 40% sensitivity [4]. When less stringent criteria are applied to define positive oximetry, the sensitivity improved to 86.6%. In studies of children with Down syndrome, oximetry has been shown to have similar sensitivity and specificity, with the caveat that it cannot differentiate obstructive versus central apnea events [6]. Additionally, the results of oximetry studies may be less reliable in obese children [7]. In a recent prospective study with 268 untreated children, a quarter of whom were obese, a majority of patients with an AHI > 5 had McGill scores > 1 [8]. However, 65% of children with moderate to severe OSAS had negative or inconclusive McGill scores, which led to their incorrect classification as mild OSAS or primary snoring. However, the oximetry studies did not overestimate disease severity in any of the patients. Overall, positive home oximetry studies are useful to establish the presence of OSAS, but caution should be used when interpreting negative or inconclusive studies since there is a significant risk of false negative results and underestimation of disease severity.

### Peripheral Arterial Tonometry

PAT is a portable, wrist-worn device, which combines a pulse oximeter, pulse rate monitor, probe for measuring arterial tone, and actigraphy. The PAT probe uses this information to monitor changes in sympathetic nervous system activation and uses an algorithm to measure respiratory disturbances. In adults, PAT strongly correlates with AHI, RDI, and sleep time as measured on PSG [9]. There are few studies using this technology in children although the device is FDA-approved

for children 12 and older. A recent prospective study involving 36 children (aged 8–15 years) demonstrated good correlation of the AHI and ODI between PSG and PAT, but reported a consistently lower AHI using PAT; this may reflect the fact that an adult-sized probe was used along with algorithms created for adults [10]. When curves were constructed to diagnose severe OSAS, the greatest diagnostic accuracy was seen at an AHI cutoff of greater than 3.5 events/hour, with a 77% sensitivity and 78% specificity. When the PAT-reported AHI was over 10 events/hour, there was 91% specificity for diagnosing severe OSAS when compared with the PSG [10].

### Drug-Induced Sleep Endoscopy

DISE involves the passage of a flexible endoscope or bronchoscope through the nose and upper airway during a pharmacologically-induced sleep-like state. This procedure allows for the visualization of airway dynamics and identification of sites of obstruction, particularly those that may occur exclusively during sleep. Maneuvers such as jaw thrust are typically performed to enhance the evaluation of obstructive sites, especially for the tongue base. DISE has been successfully used to determine surgical management options for infants with OSAS or for older children with persistent OSAS after adenotonsillectomy [11–13]. Manickam et al. reported that DISE is fairly reliable in identifying at least one site of obstruction, with reported rates between 52% and 100% in non-syndromic patients and 100% in patients with Down syndrome [14]. It is particularly helpful for evaluating the nasal cavity and laryngeal obstruction, and it allows for evaluation of the lower airway if warranted. Over 21 different scoring systems have been developed to help describe DISE findings in adults and children with the two most common being the VOTE (velum, oropharynx, tongue base, epiglottis) system and the Pringle and Croft classification [15]. However, neither system includes nasal cavity, nasopharyngeal, or laryngeal sites of obstruction, which can be significant areas of obstruction in children. Subsequent grading systems by Chan et al. [16] including the Sleep Endoscopy Rating Scale (SERS) by Lam et al. [17] have been tailored for children by including the nasal cavity, nasopharynx, and supraglottis as part of the scoring. These methods demonstrated substantial inter-/intra-rater reliability and good correlation with polysomnography (PSG) indices, particularly AHI and oxygen saturation nadir [18]. In the senior author's opinion, the advantage of DISE over cine MRI is the improved assessment of the nasal cavity and larynx.

### Cine MRI

Cine MRI provides high-resolution anatomic information and captures the dynamic movement of the upper airway while avoiding the radiation exposure involved with CT or fluoroscopy [19]. Similar to DISE, cine MRI is performed in a drug-induced

sleep-like state. However, one advantage of cine MRI when compared with DISE is the ability to simultaneously assess multiple levels of obstruction [14•]; this is particularly helpful for children with Down syndrome or neurologic issues, where multilevel obstruction is common [20, 21]. Relatively objective measurements of the soft tissues can be taken, which allows for quantification of factors such as the degree of lingual tonsil hypertrophy, soft palate collapse, or tongue base collapse. Much of the published cine MRI data involves the assessment of children with Down syndrome with OSAS, thus it is difficult to extrapolate the utility of this modality to other populations. Currently, this modality is not widely used due to limited availability at only a few highly specialized centers.

## Management of Pediatric OSA

In most pediatric cases of primary OSA, upper airway obstruction during sleep is associated with adenotonsillar hypertrophy. Accordingly, adenotonsillectomy is commonly the first line therapy in pediatric OSA. For children who are not candidates for or refuse surgical therapy, pharmacologic therapy with intranasal steroids or leukotriene receptor antagonists, or continuous positive airway pressure (CPAP) may be recommended depending on the severity of disease [22]. For children and adolescents with persistent or recurrent OSA after adenotonsillectomy, CPAP is the mainstay of treatment [23]; however, given concerns over poor compliance with CPAP therapy, various medical, dental, and surgical options may also be considered. The therapeutic strategy employed is largely dependent on the pattern and severity of persistent or recurrent OSA determined by PSG, the anatomic sites of obstruction determined by DISE and/or airway imaging, and individual patient factors, including underlying syndromic disorders or medical comorbidities.

### Pharmacologic Therapy

Intranasal steroids are effective for children with mild OSA for whom adenotonsillectomy is contraindicated or for children with mild persistent OSA after initial adenotonsillectomy. In 2001, a randomized, placebo-controlled trial evaluated the effectiveness of intranasal fluticasone in 25 children with OSA and adenotonsillar hypertrophy. For patients in the treatment arm, there was a statistically significant reduction in the mean mixed/obstructive apnea/hypnea index from  $10.7 \pm 2.6$  to  $5.8 \pm 2.2$  events/hour; patients in the placebo arm demonstrated an increase in the mean mixed/obstructive apnea/hypopnea index from  $10.9 \pm 2.3$  to  $13.1 \pm 3.6$ , ( $p = 0.04$ ) [24]. Similarly, a 2015 randomized, placebo-controlled trial evaluated intranasal mometasone in 50 children with mild OSA not previously treated with adenotonsillectomy. The mean oAHI decreased from  $2.7 \pm 0.2$  to  $1.7 \pm 0.3$  events/

hour in the treatment group and increased from  $2.5 \pm 0.2$  to  $2.9 \pm 0.6$  in the placebo group, ( $p = 0.039$ ) [25•].

Several studies have evaluated the effectiveness of montelukast, a leukotriene modifier, in the treatment of children with OSA. One randomized controlled trial compared daily oral montelukast to placebo in 46 children with mild to moderate OSA who had not previously undergone adenotonsillectomy. In the montelukast group, there was a significant reduction in the obstructive apnea index (OAI), adenoid size, and parent-reported symptoms despite no significant reduction in mean oAHI after treatment ( $6.0 \pm 3.2$  to  $3.6 \pm 2.3$  events/hour,  $p = 0.07$ ) [26]. Likewise, a significant improvement in OSA severity was demonstrated among 57 children who had not previously undergone adenotonsillectomy in a randomized, placebo-controlled trial of montelukast. Twenty of 28 children receiving daily oral montelukast had an improvement in AHI, and the mean AHI was reduced from  $9.2 \pm 4.1$  pre-treatment to  $4.2 \pm 2.8$  post-treatment ( $p < 0.0001$ ). The mean oxygen saturation nadir was also significantly improved. These same parameters were unchanged in the placebo-controlled group [27•]. These studies suggest that the administration of intranasal steroids or leukotriene modifiers in children with OSA who have not undergone adenotonsillectomy is associated with a significant reduction in the severity of disease by PSG, although the long-term follow-up and clinical sequelae of disease were not specifically addressed; complete resolution of disease is not necessarily assured.

A randomized prospective study compared oral montelukast to observation in 58 children with mild persistent OSA after adenotonsillectomy. The authors reported a significant improvement in the mean AHI ( $3.55 \pm 1.15$  to  $2.20 \pm 0.93$ ,  $p < 0.001$ ), oxygen saturation nadir ( $83.72 \pm 6.75$  to  $89.41 \pm 4.81\%$ ,  $p < 0.001$ ), and validated Pediatric Sleep Questionnaire (PSQ) score ( $0.39 \pm 0.11$  to  $0.27 \pm 0.09$ ,  $p < 0.001$ ) [28•]. Finally, combination therapy with intranasal steroids and oral leukotriene modifiers was evaluated in children with residual mild OSA after adenotonsillectomy. A significant reduction in AHI ( $3.9 \pm 1.2$  to  $0.3 \pm 0.3$  events/hour) was demonstrated in 22 children treated with intranasal budesonide and oral montelukast after initial adenotonsillectomy [29]. These studies suggest that leukotriene modifiers and intranasal steroids, alone or in combination, are effective adjuvants for mild residual OSA.

### Non-pharmacologic Therapy

For children with OSA who are not candidates for adenotonsillectomy or who have residual disease after initial adenotonsillectomy, CPAP is a safe and effective treatment option. By providing constant positive pressure, CPAP corrects partial and complete upper airway obstruction during

sleep. With consistent use, CPAP therapy has been correlated with improvements in AHI, daytime behavior, daytime sleepiness, and blood pressure [30, 31]. However, adherence rates to CPAP therapy vary widely (33–98%), resulting in a large number of children with OSA who are untreated due to poor compliance or inconsistent use [30, 32, 33].

Treatment with nasal insufflation (TNI) via high-flow nasal cannula (HFNC) may be an effective alternative to CPAP therapy in these children [34]. HFNC provides non-invasive respiratory support with the delivery of humidified and heated oxygen at flow rates exceeding that of normal inspiratory flow, thus generating positive airway pressure and reducing upper airway collapse. A case series of five children with moderate to severe OSA treated with HFNC, either in hospital or at home, showed a significant reduction in the mean AHI ( $22.9 \pm 15.2$  to  $5.0 \pm 2.8$ ,  $p = 0.034$ ) and a significant improvement in the mean oxygen saturation nadir ( $65.0 \pm 11.97$  to  $81.4 \pm 6.43\%$ ,  $p = 0.011$ ) [35]. Subsequently, in a study of 10 children with moderate to severe OSA treated with HFNC, there were significant improvements in median oAHI from 11.1 (interquartile range (IQR) 8.7–18.8) to 2.1 (IQR 1.7–2.2) events/hour ( $p = 0.002$ ) and mean oxygen saturation nadir from 76.0% (IQR 67.3–82.3) to 79.5% (IQR 77.2–86.0) ( $p = 0.032$ ). Similar improvements were achieved in patients who were overweight/obese versus non-overweight/obese [36]. Further randomized studies comparing CPAP and HFNC and evaluating long-term effectiveness and compliance are needed.

### Orthodontic and Orthognathic Therapy

Various craniofacial abnormalities are associated with OSA in children and adolescents. A narrow, high-arched palate decreases nasal cavity volume, increases nasal airway resistance, and predisposes children to mouth breathing. Mandibular retropositioning due to mandibular insufficiency or delayed mandibular growth is associated with posterior displacement of the tongue, often resulting in tongue base obstruction [37]. In select patients, orthodontic treatment and orthognathic surgery improve dental alignment and maxillomandibular position in order to enlarge the airway and correct intranasal, velopharyngeal, and/or retroglossal obstruction contributing to OSA [38].

Intraoral appliances are designed to stabilize the tongue in an anterior position or actively protrude the mandible forward in order to relieve tongue base obstruction during sleep [39]. Additionally, devices may stimulate the upper airway dilatory muscles, thus reducing airway collapsibility [40]. In one study, 19/32 children with mild to severe OSA ( $AHI > 2$ ) and dental malocclusion were randomly assigned to a 6-month trial of a mandibular and tongue positioning device, which was worn continuously. After orthodontic therapy, there was a significant reduction in mean AHI ( $7.1 \pm 4.6$  to  $2.6 \pm 2.2$  events/hour,  $p < 0.001$ ). 62.4% had an AHI reduction of at least 50%, and parent-reported respiratory and daytime

symptoms improved in over half of patients [41]. A systematic review, including three additional retrospective studies evaluating the effectiveness of mandibular advancement devices, with or without a tongue retainer, reported that these appliances appear to significantly reduce AHI when OSA is associated with mandibular retrognathia; however, mandibular advancement appliances alone have not been shown to completely correct OSA. The authors of the review concluded that the limited evidence suggests short-term improvements of AHI but does not demonstrate its effectiveness in the treatment of pediatric OSA and data regarding compliance with therapy and long-term follow-up is lacking [42].

Mandibular advancement and mandibular distraction osteogenesis surgeries are employed in the treatment of children with underlying mandibular insufficiency with associated OSA; this condition is typically seen in syndromic patients with Pierre Robin sequence, Treacher Collins syndrome, Stickler syndrome, and velocardiofacial syndrome. A 2018 systematic review and meta-analysis evaluated mandibular advancement for the treatment of pediatric OSA, including 37 studies with 376 patients. After surgical therapy, there was a reduction in mean AHI from  $41.1 \pm 35.8$  to  $4.5 \pm 6.0$  events/hour, and the mean oxygen saturation nadir increased from  $76.8 \pm 13.0$  to  $91.1 \pm 8.6$ . Among 105 patients for whom individual patient data could be analyzed, surgical success ( $> 50\%$  reduction and post-treatment  $AHI < 5$ ) was achieved in 73.4%, and surgical cure (post-treatment  $AHI \leq 1$ ) was achieved in 25.5%. The authors concluded that mandibular advancement surgery dramatically improves OSA in select children with retrognathia and mandibular insufficiency [43].

Rapid maxillary expansion (RME) is an orthodontic treatment that corrects dental crowding in children with maxillary skeletal constriction by increasing the transverse upper dental arch size and widening the palate. As a result, the nasal cavity volume is increased and the tongue position is normalized [44]. A 2017 systematic review and meta-analysis evaluated outcomes of RME in children with OSA. The authors analyzed 17 studies including 314 children with high-arched and/or narrowed palates and OSA who underwent RME. For children with less than 3 years of follow-up after RME, the mean AHI decreased from  $8.9 \pm 7.0$  to  $2.7 \pm 3.3$  events/hour. For 52 children who had follow-up greater than 3 years, the mean AHI decreased from  $7.1 \pm 5.7$  to  $1.5 \pm 1.8$  events/hour ( $p < 0.001$ ). Overall, there was a 70% reduction in reported AHI; among 90 children for whom individual data was analyzed, the cure rate was 25.6% [45]. RME may be considered in select children with maxillary skeletal constriction and OSA, with greater improvements anticipated in those with no tonsillar hypertrophy or with residual OSA following prior adenotonsillectomy. However, long-term follow-up in randomized treatment and control group patients is needed in order to determine the effectiveness of RME versus maxillary growth from age alone.

Finally, a retrospective study published in 2017 evaluated the effectiveness of bimaxillary expansion (BE) as a treatment option for pediatric OSA. Thirty of 45 children (66%) with mild to severe OSA who had not previously undergone adenotonsillectomy showed improvement in AHI at 3–6 months after treatment with BE; however, effect sizes were modest, with reduction in pre-treatment median AHI of 7.6 (range 0.1–32.5) to post-treatment median AHI of 6.7 (0.0–30.8) events/hour ( $p = 0.05$ ) [46].

## Surgical Treatment for Pediatric OSA

### Lateral Pharyngoplasty and Expansion Sphincter Pharyngoplasty

Lateral pharyngeal wall collapse is a potential site of upper airway obstruction in both children and adults with OSA. In an effort to improve cure rates of primary OSA in children undergoing adenotonsillectomy, treatment with concomitant lateral pharyngoplasty with suture closure of the tonsillar pillars had previously been considered; however, prior pediatric studies did not demonstrate improved effectiveness in comparison with adenotonsillectomy alone [47, 48].

Expansion sphincter pharyngoplasty is used to reduce lateral pharyngeal wall collapse and combines tonsillectomy with the rotation of the palatopharyngeus muscle along a superior-lateral vector; it may be combined with partial uvulectomy and/or closure of the tonsillar pillars. In a retrospective review of 25 children with AHI > 10 and lateral pharyngeal wall collapse identified by pre-operative DISE, significant improvement in the postoperative mean AHI (from  $60.5 \pm 38.5$  to  $2.0 \pm 3.9$  events/hour) was demonstrated after tonsillectomy with modified expansion sphincter pharyngoplasty. The overall cure rate (AHI < 1) was 64%. Both the postoperative AHI and cure rate were significantly better among children who underwent modified expansion sphincter pharyngoplasty compared with AHI-matched children who underwent adenotonsillectomy alone ( $p < 0.05$ ) [49].

### Lingual Tonsillectomy

Lingual tonsil hypertrophy may result in prolapse of the posterior tongue and subsequent obstruction of the retroglottal airway. It can be identified on awake flexible nasopharyngoscopy, DISE, and cine MRI. However, lingual tonsillar size is often overestimated on endoscopic exam alone, when enlargement or repositioning of the underlying base of tongue results in further prolapse of lingual tonsillar tissue.

Removal of the lingual tonsils is the most common surgery for persistent OSA after adenotonsillectomy. Lingual tonsil tissue is frequently removed transorally with radiofrequency ablation, suction cautery, or microdebrider. Particular attention is turned to clearing the vallecular space, where residual tissue

is most likely to prolapse into the airway. Lingual tonsillectomy is generally safe, with complications rates comparable to those of adenotonsillectomy [50]. In a 2016 systematic review, lingual tonsillectomy performed among 141 children overall resulted in an improvement in mean AHI from 13.9 to 8.0 events/hour [14]. In two studies, postoperative AHI < 5 was achieved in 57–61% of patients, and postoperative AHI < 1 was achieved in 17–22% of patients [51, 52]. For children with persistent OSA after tonsillectomy, lingual tonsillectomy may produce a significant improvement in AHI when lingual tonsillar hypertrophy is identified.

### Posterior Midline Glossectomy

Posterior midline glossectomy (PMG) is employed in children who have predominant tongue base obstruction from relative macroglossia or retroposition of the posterior tongue and is frequently combined with lingual tonsillectomy. With PMG, a wedge of posterior tongue tissue is ablated or excised via open or submucosal approaches, most commonly using radiofrequency ablation. The resultant scar formation along the posterior aspect of the tongue may result in further improvement in the surgical outcome.

In a study of 13 children with Down syndrome and OSA after adenotonsillectomy who underwent PMG with lingual tonsillectomy, the mean AHI improved from 47 to 5.6 events/hour in normal to overweight patients; however, similar improvements in AHI were not demonstrated in obese patients [53]. In an additional study of 26 patients undergoing DISE-directed multilevel surgery, 16 (62%) had significant improvements in AHI following PMG [13].

### Tongue Suspension

Tongue suspension is another technique directed at relieving base of tongue collapse, particularly in patients who demonstrate glossoptosis of the superior aspect of the posterior base of the tongue on cine MRI. In this procedure, the base of the tongue is suspended anteriorly to the genial tubercle of the mandible with a non-absorbable suture passed through and around the soft tissue at the base of the tongue. The procedure may be combined with partial midline glossectomy in order to further reduce the bulk and collapsibility of the tongue base through thermally-induced fibrosis.

In a study of 31 patients with OSA and tongue base obstruction undergoing tongue suspension with concurrent radiofrequency ablation of the tongue base, the mean AHI improved from 14.1 to 6.4 events/hour, and mean oxygen saturation nadir improved from 87 to 91%. Overall, the success rate (AHI < 5 and oxygen saturation nadir > 90%) was 61% [54].

## Hypoglossal Nerve Stimulation

Hypoglossal nerve stimulators are implantable devices that stimulate the anterior branches of the hypoglossal nerve to alleviate tongue base obstruction during inspiration. In 2014, the Food and Drug Administration (FDA) approved hypoglossal nerve stimulator implantation for the treatment of moderate to severe OSA in adults, with subsequent studies showing both success and compliance with long-term use [55, 56].

To date, pediatric hypoglossal nerve stimulation has been limited to studies in adolescents with Down syndrome who have persistent moderate to severe OSA and are unable to tolerate CPAP [57, 58•]. In a case series of six adolescents with Down syndrome and AHI > 10 implanted with the hypoglossal nerve stimulator, significant improvements in AHI were seen. After at least 6 months of therapy, a 56–85% reduction in AHI was demonstrated, with AHI < 5 in four of six patients [58•]. This suggests that hypoglossal nerve stimulator implantation is a potential therapeutic option for children with persistent severe OSA, but further research is needed.

## Hyoid Suspension

In patients with OSA, the hyoid bone tends to sit lower in the neck than in those without OSA, which may lead to the increased posterior collapse of soft tissues and resultant hypopharyngeal airway obstruction. In order to elevate the hyoid, suspension sutures may be placed around the midportion of the hyoid bone and secured to either the thyroid cartilage or the lower anterior mandible. The hyoid bone is suspended anteriorly, resulting in increased posterior airway spaced and improvement in hypopharyngeal obstruction [59].

A review of the effectiveness of hyoid suspension surgeries for the treatment of OSA in adults [59] reported significant improvements in mean AHI and Epworth Sleepiness Scale scores [59, 60]. However, there is a paucity of published data regarding hyoid suspension in children. Given the demonstrable benefits in adult OSA, further investigation into the use and effectiveness of hyoid suspension for persistent OSA in children is necessary.

## Supraglottoplasty

Infants and children with severe laryngomalacia may experience significant OSA (reported in 3%), and supraglottoplasty remains the mainstay of treatment for these infants. Supraglottoplasty typically involves surgery to address foreshortened aryepiglottic folds, epiglottic prolapse, and/or arytenoid prolapse. Two meta-analyses reported significant improvements in mean AHI and oxygen saturation nadir. Across four included studies, the mean difference in AHI after supraglottoplasty was  $-12.5$  events/hour (95% CI,  $-21.14$  to  $-3.78$ ,  $p = 0.005$ ) [61•]. In an additional six studies, the mean

difference in AHI was  $-10.7$  events/hour (95% CI,  $-14.9$  to  $-6.5$ ) (Camacho et al. 2016). Surgical cure of OSA with supraglottoplasty was reported in 11–27% of cases, when individual patient data was available for analysis [61•, 62•].

## Conclusions

While polysomnography remains the gold standard for diagnosis of OSA in children, reliable alternative diagnostic modalities are needed due to the limitations associated with cost, access, and availability. Overnight pulse oximetry is widely available, relatively low cost, and easy to analyze with formal scoring systems; positive home oximetry studies are useful to establish the presence of OSA, but disease severity may be underestimated. DISE and cine MRI may allow for more accurate diagnosis of anatomic sites of upper airway obstruction contributing to OSA and direct therapy beyond adenotonsillectomy in children who have residual or persistent disease. Pharmacologic therapy with intranasal steroids and oral leukotriene inhibitors has been shown to be effective for the treatment of mild primary OSA or mild residual OSA after adenotonsillectomy. For children and adolescents with persistent or recurrent OSA after adenotonsillectomy, CPAP remains the mainstay of treatment; however, various medical, dental, and surgical options may be considered for children who are intolerant of or noncompliant with CPAP therapy. The effectiveness of therapy is dependent on the severity of disease, anatomic sites of obstruction, and individual patient factors, including underlying syndromic disorders or medical comorbidities. Further study is required to evaluate the long-term effectiveness of treatment.

## Compliance with Ethical Standards

**Conflict of Interest** Philip D. Knollman and Aimee A. Kennedy declare that they have no conflict of interest. Dr. Ishman reports consulting fees from Genus Life Science.

**Human and Animal Rights and Informed Consent** This article does not contain any studies with human or animal subjects performed by any of the authors.

## References

Papers of particular interest, published recently, have been highlighted as:

- Of importance
1. Bhattacharjee R, Kheirandish-Gozal L, Spruyt K, Mitchell RB, Promchiarak J, Simakajornboon N, et al. Adenotonsillectomy outcomes in treatment of obstructive sleep apnea in children: a multi

- center retrospective study. *Am J Respir Crit Care Med.* 2010;182(5):676–83.
2. Friedman M, Wilson M, Hsin-Ching L, Chang HW. Updated systematic review of tonsillectomy and adenoidectomy for treatment of pediatric obstructive sleep apnea/hypopnea syndrome. *Otolaryngol Head Neck Surg.* 2009;140(6):800–8.
  3. Mitchell RB, Garetz S, Moore RH, Rosen CL, Marcus CL, Katz ES, et al. The use of clinical parameters to predict obstructive sleep apnea syndrome severity in children: the Childhood Adenotonsillectomy (CHAT) study randomized clinical trial. *JAMA Otolaryngol Head Neck Surg.* 2015;141(2):130–6.
  4. Kaditis A, Kheirandish-Gozal L, Gozal D. Pediatric OSAS: oximetry can provide answers when polysomnography is not available. *Sleep Med Rev.* 2016;27:96–105.
  5. Nixon GM, Kermack AS, David GM, et al. Planning adenotonsillectomy in children with obstructive sleep apnea: the role of overnight oximetry. *Pediatrics.* 2004;113:19–25.
  6. Coverstone AM, Bired M, Sicard M, et al. Overnight pulse oximetry for evaluation of sleep apnea among children with trisomy 21. *J Clin Sleep Med.* 2014;10(12):1309–15.
  7. Eyck AV, Lambrechts C, Vanheeswijck L, et al. The role of nocturnal pulse oximetry in the screening for obstructive sleep apnea in obese children and adolescents. *Sleep Med.* 2015;16:1409–12.
  8. Villa MP, Pietropaoli N, Supino MC, et al. Diagnosis of pediatric obstructive sleep apnea syndrome in settings with limited resources. *JAMA Otolaryngol Head Neck Surg.* 2015;141(11):990–6.
  9. Yalamanchali S, Farajian V, Hamilton C, Pott TR, Samuelson CG, Friedman M. Diagnosis of obstructive sleep apnea by peripheral arterial tonometry: meta-analysis. *JAMA Otolaryngol Head Neck Surg.* 2013;139(12):1343–50.
  10. Tanphairchitr A, Thianboonsong A, Banhiran W, et al. Watch peripheral arterial tonometry in the diagnosis of pediatric obstructive sleep apnea. *Otolaryngol Head Neck Surg.* 2018;159(1):166–72.
  11. Boudewyns A, Van de Heyning P, Verhulst S. Drug-induced sedation endoscopy in children <2 years with obstructive sleep apnea syndrome: upper airway findings and treatment outcomes. *Eur Arch Otorhinolaryngol.* 2017;274:2319–25.
  12. He S, Peddireddy NS, Smith DF, Duggins AL, Heubi C, Shott SR, et al. Outcomes of drug-induced sleep endoscopy-directed surgery for pediatric obstructive sleep apnea. *Otolaryngol Head Neck Surg.* 2018;158(3):559–65.
  13. Wooten CT, Chinnadurai S, Goudy S. Beyond adenotonsillectomy: outcomes of sleep endoscopy-directed treatments in pediatric obstructive sleep apnea. *Int J Pediatr Otorhinolaryngol.* 2014;78:1158–62.
  14. Manickam PV, Shott SR, Boss EF, et al. Systematic review of site of obstruction identification and non-CPAP treatment options for children with persistent pediatric obstructive sleep apnea. *Laryngoscope.* 2016;126(2):491–500. **This is a systematic review evaluating the effectiveness of lingual tonsillectomy, demonstrating an improvement in mean AHI from 13.9 to 8.0 events/hour. In two included studies, postoperative AHI < 5 was achieved in 57–61% of patients, and postoperative AHI < 1 was achieved in 17–22% of patients.**
  15. Amos JM, Durr ML, Nardone HC, Baldassari CM, Duggins A, Ishman SL. Systematic review of drug-induced sleep endoscopy scoring systems. *Otolaryngol Head Neck Surg.* 2018;158(2):240–8.
  16. Chan DK, Liming BJ, Horn DL, Parik SR. A new scoring system for upper airway pediatric sleep endoscopy. *JAMA Otolaryngol Head Neck Surg.* 2014;140(7):595–602.
  17. Lam DJ, Weaver EM, Macarthur CJ, Milczuk HA, O'Neill E, Smith TL, et al. Assessment of pediatric obstructive sleep apnea using a drug-induced sleep endoscopy rating scale. *Laryngoscope.* 2016;126:1492–8.
  18. Dahl JP, Miller C, Purcell PL, Zopf DA, Johnson K, Horn DL, et al. Airway obstruction during drug-induced sleep endoscopy correlates with apnea-apnea-hypopnea index and oxygen nadir in children. *Otolaryngol Head Neck Surg.* 2016;155(4):676–80.
  19. Donnelly L. Obstructive sleep apnea in pediatric patients: evaluation with cine MR sleep studies. *Radiology.* 2005;236(3):768–78.
  20. Donnelly LF, Shott SR, LaRose CR, et al. Causes of persistent obstructive sleep apnea despite previous tonsillectomy and adenoidectomy in children with down syndrome as depicted on static and dynamic cine MRI. *Am J Roentgenol.* 2004;183(1):175–81.
  21. Isaiah A, Kiss E, Olomu P, Koral K, Mitchell RB. Characterization of upper airway obstruction using cine MRI in children with residual obstructive sleep apnea after adenotonsillectomy. *Sleep Med.* 2018;50:79–86.
  22. Economidou NT, Ferini-Strambi L, Steiropoulos P. Sleep-related drug therapy in special conditions: children. *Sleep Med Clin.* 2018;13:251–62.
  23. Marcus CL, Rosen G, Ward SL. Adherence to and effectiveness of positive airway pressure therapy in children with obstructive sleep apnea. *Pediatrics.* 2006;117:442–51.
  24. Brouillette RT, Manoukian JJ, Ducharme FM, Oudjhane K, Earle LG, Ladan S, et al. Efficacy of fluticasone nasal spray for pediatric obstructive sleep apnea. *J Pediatr.* 2001;138(6):838–44.
  25. Chan CCK, Au CT, Lam HS, et al. Intranasal corticosteroids for mild childhood obstructive sleep apnea – randomized, placebo-controlled study. *Sleep Med.* 2015;16:358–63. **This is a randomized, placebo-controlled trial evaluating intranasal mometasone administered over 16 weeks in 50 children with mild OSA (AHI  $\geq 1-5$ ) not previously treated with adenotonsillectomy. The mean AHI decreased from  $2.7 \pm 0.2$  to  $1.7 \pm 0.3$  events/hour in the treatment group and increased from  $2.5 \pm 0.2$  to  $2.9 \pm 0.6$  in the placebo group, ( $p = 0.039$ ).**
  26. Goldbart AD, Greenberg-Dotan S, Tal A. Montelukast for children with obstructive sleep apnea: a double-blind, placebo-controlled study. *Pediatrics.* 2012;130(3):e575–80.
  27. Kheirandish-Gozal L, HPR B, Gozal D. Montelukast for children with obstructive sleep apnea: results of a double-blind, randomized, placebo-controlled trial. *Ann Am Thorac Soc.* 2016;13(10):1736–41. **This is a randomized, placebo-controlled study of 57 children not previously treated with adenotonsillectomy treated with a 16-week trial of montelukast. In the treatment group, 20/28 children receiving daily oral montelukast had improvement in AHI, and the mean AHI was reduced from  $9.2 \pm 4.1$  pre-treatment to  $4.2 \pm 2.8$  post-treatment ( $p < 0.0001$ ).**
  28. Wang B, Liang J. The effect of montelukast on mild persistent OSA after adenotonsillectomy in children: a preliminary study. *Otolaryngol Head Neck Surg.* 2017;156(5):952–4. **This is a randomized prospective study comparing oral montelukast to observation in 58 children with mild persistent OSA after adenotonsillectomy, demonstrating a significant improvement in the mean AHI ( $3.55 \pm 1.15$  to  $2.20 \pm 0.93$ ,  $p < 0.001$ ), oxygen saturation nadir ( $83.72 \pm 6.75$  to  $89.41 \pm 4.81\%$ ,  $p < 0.001$ ), and validated Pediatric Sleep Questionnaire (PSQ) score ( $0.39 \pm 0.11$  to  $0.27 \pm 0.09$ ,  $p < 0.001$ ) after a 12-week course of treatment.**
  29. Kheirandish L, Goldbart AD, Gozal D. Intranasal steroids and oral leukotriene modifier therapy in residual sleep-disordered breathing after tonsillectomy and adenoidectomy in children. *Pediatrics.* 2006;117(1):e61–6.
  30. Ramirez A, Khirani S, Aloui S, Delord V, Borel JC, Pépin JL, et al. Continuous positive airway pressure and noninvasive ventilation adherence in children. *Sleep Med.* 2013;14:1290–4.
  31. DelRosso LM, King J, Ferri R. Systolic blood pressure elevation in children with obstructive sleep apnea is improved with positive airway pressure use. *J Pediatr.* 2018;195:102–7.
  32. Xanthopoulos MS, Kim JY, Blechner M, et al. Self-efficacy and short-term adherence to continuous positive airway pressure treatment in children. *Sleep.* 2017;40(7):1–7.

33. Mihai R, Vandeleur M, Pecoraro S, Davey MJ, Nixon GM. Autotitrating CPAP as a tool for CPAP initiation in children. *J Clin Sleep Med*. 2017;13(5):713–9.
34. McGinley B, Halbower A, Schwartz AR, Smith PL, Patil SP, Schneider H. Effect of high-flow open nasal cannula system on obstructive sleep apnea in children. *Pediatr*. 2009;124(1):179–88.
35. Joseph L, Goldberg S, Shitrit M, Picard E. High-flow nasal cannula therapy for obstructive sleep apnea in children. *J Clin Sleep Med*. 2015;11(9):1007–10.
36. Hawkins S, Huston S, Campbell K, et al. High-flow, heated, humidified air via nasal cannula treats CPAP-intolerant children with obstructive sleep apnea. *J Clin Sleep Med*. 2017;13(8):981–9. **This is a study of 10 children with moderate to severe OSA treated with HFNC, demonstrating significant improvements in median AHI from 11.1 (interquartile range (IQR) 8.7–18.8) to 2.1 (IQR 1.7–2.2) events/hour ( $p = 0.002$ ) and mean oxygen saturation nadir from 76.0% (IQR 67.3–82.3) to 79.5% (IQR 77.2–86.0) ( $p = 0.032$ ). Two of 10 patients demonstrated resolution of OSA.**
37. Rohra AK, Demko CA, Hans MG, et al. Sleep disordered breathing in children seeking orthodontic care. *Am J Orthod Dentofac Orthop*. 2018;154(1):65–71.
38. Naran S, Steinbacher DM, Taylor JA. Current concepts in orthognathic surgery. *Plast Reconstr Surg*. 2018;141(6):925–36.
39. Conley RS. Management of sleep apnea: a critical look at intra-oral appliances. *Orthod Craniofac Res*. 2015;18(1):83–90.
40. Ferguson KA, Cartwright R, Rogers R, Schmidt-Nowara W. Oral appliances for snoring and obstructive sleep apnea: a review. *Sleep*. 2006;29:244–62.
41. Villa MP, Bernkopf E, Pagani J, et al. Randomized controlled study of an oral jaw-positioning appliance for the treatment of obstructive sleep apnea in children with malocclusion. *Am J Respir Crit Care Med*. 2002;165:123–7.
42. Nazarali N, Altalibi M, Nazarali S, et al. Mandibular advancement appliances for the treatment of paediatric obstructive sleep apnea: a systematic review. *Eur J Orthod*. 37(6):618–26. **This is a systematic review evaluating the effectiveness of mandibular advancement devices in the treatment of OSA, reporting a significant reduction in AHI when OSA is associated with mandibular retrognathia; however, mandibular advancement appliances alone have not been shown to completely correct OSA.**
43. Noller MW, Guilleminault C, Gouveia CJ, Mack D, Neighbors CL, Zaghi S, et al. Mandibular advancement for pediatric obstructive sleep apnea: a systematic review and meta-analysis. *J Craniomaxillofac Surg*. 2018;46:1296–302.
44. Machado AJ, Crespo AN, Pauna HF. Rapid maxillary expansion in pediatric patients with obstructive sleep apnea: current and future perspectives. *Sleep Med*. 2018;51:7–8.
45. Camacho M, Chang ET, Song SA. Rapid maxillary expansion for pediatric obstructive sleep apnea: a systematic review of meta-analysis. *Laryngoscope*. 2017;127:1712–9. **This is a systematic review and meta-analysis evaluating outcomes of rapid maxillary expansion in children with OSA and high-arched and/or narrowed palate. For 52 children who had follow-up greater than 3 years, the mean AHI decreased from  $7.1 \pm 5.7$  to  $1.5 \pm 1.8$  events/hour ( $p < 0.001$ ). Overall, there was a 70% reduction in reported AHI; the cure rate was 25.6%.**
46. Quo SD, Hyunh N, Guilleminault C. Bimaxillary expansion therapy for pediatric sleep-disordered breathing. *Sleep Med*. 2017;30:45–51.
47. Friedman M, Samuelson CG, Hamilton C, Maley A, Taylor D, Kelley K, et al. Modified adenotonsillectomy to improve cure rates for pediatric obstructive sleep apnea: a randomized controlled trial. *Otolaryngol Head Neck Surg*. 2012;147(1):132–8.
48. Merrell JA, Shott SR. OSAS in down syndrome: T&A versus T&A plus lateral pharyngoplasty. *Int J Pediatr Otorhinolaryngol*. 2007;71:1197–203.
49. Ulualp SO. Modified expansion sphincter pharyngoplasty for treatment of children with obstructive sleep apnea. *JAMA Otolaryngol Head Neck Surg*. 2014;140(9):817–22.
50. DeMarcantonio MA, Sencer E, Meitzen-Derr J, et al. The safety and efficacy of pediatric lingual tonsillectomy. *Int J Pediatr Otorhinolaryngol*. 2016;91:6–10.
51. Chan DK, Jan TA, Koltai PJ. Effect of obesity and medical comorbidities on outcomes after adjunct surgery for obstructive sleep apnea in cases of adenotonsillectomy failure. *Arch Otolaryngol Head Neck Surg*. 2012;138:891–6.
52. Lin AC, Koltai PJ. Persistent pediatric obstructive sleep apnea and lingual tonsillectomy. *Arch Otolaryngol Head Neck Surg*. 2009;141:81–5.
53. Probst EJ, Amin R, Talwar N, et al. Midline posterior glossectomy and lingual tonsillectomy in obese and nonobese children with down syndrome: biomarkers for success. *Laryngoscope*. 2017;127:757–63.
54. Wooten CT, Shott SR. Evolving therapies to treat retroglossal and base-of-tongue obstruction in pediatric obstructive sleep apnea. *Arch Otolaryngol Head Neck Surg*. 2010;136(10):983–7.
55. Strollo PJ, Soose RJ, Maurer JT, et al. STAR trial group. Upper-airway stimulation for obstructive sleep apnea. *N Engl J Med*. 2014;370:139–49.
56. Woodson BT, Soose RJ, Gillespie MB, Strohl KP, Maurer JT, de Vries N, et al. Three-year outcomes of cranial nerve stimulation for obstructive sleep apnea: the STAR trial. *Otolaryngol Head Neck Surg*. 2016;154(1):181–8.
57. Diercks GR, Keamy D, Kinane TB, et al. Hypoglossal nerve stimulator implantation in an adolescent with Down syndrome and sleep apnea. *Pediatrics*. 2016;137(5):e1–5.
58. Diercks GR, Wentland C, Keamy D, et al. Hypoglossal nerve stimulation in adolescents with Down syndrome and obstructive sleep apnea. *JAMA Otolaryngol Head Neck Surg*. 2018;144(1):37–42. **This is a case series of six adolescents with Down syndrome and AHI > 10 implanted with the hypoglossal nerve stimulator showing a 56–85% reduction in AHI with at least 6 months of therapy. This suggests that hypoglossal nerve stimulator implantation is a potential therapeutic option for children with persistent severe OSA.**
59. Song SA, Wei JW, Buttram J, et al. Hyoid surgery alone for obstructive sleep apnea: a systematic review and meta-analysis. *Laryngoscope*. 2016;126:1702–8.
60. Stuck BA, Neff W, Hormann K, et al. Anatomic changes after hyoid suspension for obstructive sleep apnea; an MRI study. *Otolaryngol Head Neck Surg*. 2005;133:397–402.
61. Farhood Z, Ong AA, Nguyen SA, et al. Objective outcomes of supraglottoplasty for children with laryngomalacia and obstructive sleep apnea: a meta-analysis. *JAMA Otolaryngol Head Neck Surg*. 2016;142(7):665–71. **This is a meta-analysis evaluating supraglottoplasty in children with laryngomalacia and OSA, showing significant improvements in mean AHI ( $-12.5$  events/hour (95% CI,  $-21.14$  to  $-3.78$ ,  $p=0.005$ )) and oxygen saturation nadir.**
62. Camacho M, Dunn B, Torre C, et al. Supraglottoplasty for laryngomalacia with obstructive sleep apnea: a systematic review and meta-analysis. *Laryngoscope*. 2016;126:1246–55. **This is a meta-analysis including six studies evaluating supraglottoplasty for laryngomalacia in children with OSA, demonstrating a reduction in the mean AHI of  $-10.7$  events/hour (95% CI,  $-14.9$  to  $-6.5$ ).**