

# Obstructive sleep apnea syndrome in the pediatric age: the role of the otorhinolaryngologist

M. DE BENEDETTO<sup>1</sup>, M. ARIGLIANI<sup>1</sup>, A. BALLACCHINO<sup>2</sup>, P. CASSANO<sup>3</sup>,  
D.M. TORALDO<sup>4</sup>, M. RALLI<sup>5</sup>, F.M. PASSALI<sup>6</sup>, D. PASSALI<sup>7</sup>

<sup>1</sup>ENT Unit, Vito Fazzi Hospital, ASL Lecce, Lecce, Italy

<sup>2</sup>Qualified Expert in Respiratory Disorders in Sleep, Caltanissetta, Italy

<sup>3</sup>Department of Otolaryngology, Università degli Studi di Foggia, Foggia, Italy

<sup>4</sup>Department of Rehabilitation, Cardiorespiratory Rehabilitation Unit, ASL Lecce, Vito Fazzi Hospital, Lecce, Italy

<sup>5</sup>Department of Sense Organs, Sapienza University of Rome, Rome, Italy

<sup>6</sup>Department of Clinical Sciences and Translational Medicine, Tor Vergata University, Rome, Italy

<sup>7</sup>Department of Medicine, Surgery and Neuroscience, ENT Clinic, University of Siena, Siena, Italy

**Abstract.** – **OBJECTIVE:** Obstructive sleep apnea (OSA) is the primary indication for tonsillectomy, one of the most common pediatric surgical procedures, commonly performed in association with adenoidectomy. The objective of this review article is to evaluate the role of the otorhinolaryngologist in pediatric OSA.

**MATERIALS AND METHODS:** A literature review has been performed on the following topics: peculiarities of sleep-disordered breathing in pediatric age; discrimination of sleep disorders; adenotonsillar hypertrophy; surgical techniques; adjuvant surgical procedures.

**RESULTS:** The role of the otorhinolaryngologist in pediatric OSA is important for the evaluation of the upper airways and of essential biometric and polysomnographic data and for indication and execution of appropriate surgical treatment. In the majority of healthy children, adenotonsillectomy for OSA results in a dramatic improvement in respiratory parameters as measured by polysomnography. When post-surgical residual OSA occurs, it is essential to monitor patients by means of drug-induced sleep endoscopy (DISE).

**CONCLUSIONS:** Otolaryngologic assessment is of paramount importance to correctly classify a child with OSA. Correct inspection of the upper airway and quantification of the quality of sleep through polysomnography lead to the right therapeutic choice. Knowledge of different surgical techniques helps to deal with residual OSA after studying the obstruction sites by drug-induced sedation endoscopy.

## Key Words

Obstructive sleep apnea, Cardio-respiratory monitoring, Drug-induced sleep endoscopy.

## List of Abbreviations

OSA: obstructive sleep apnea; SDB: sleep-disordered breathing; REM: rapid eye movement; PSG: polysomnography; UARS: Upper Airway Resistance Syndrome; DISE: drug-induced sedation endoscopy; AHI: apnea hypopnea index; VOTE: Velum oropharynx tongue base epiglottis; NOHL: Nose Oropharynx Hypopharynx and Larynx; post-AT: after adenotonsillectomy.

## Introduction

In the management of the child with obstructive obstructive sleep apnea (OSA) and, more broadly, sleep-disordered breathing (SDB), the role of the otorhinolaryngologist is to provide the right indication and execution of the surgical procedure. In choosing the surgical procedure, possible complication factors must always be evaluated. These do not usually occur during the execution of the surgery; risk factors identified in the preoperative phase help predict likely postoperative complications, and thus inform surgical strategy<sup>1</sup>. Genesis of SDB is multifactorial. Among the most common causes, we consider adenotonsillar hypertrophy (45-50%) and obesity (35-45%) as being the main causes for a reduction in pharynx size. Furthermore, obesity also determines a reduction in the tone of the muscles located in the lateral walls of the pharynx. Other causes may include craniofacial malformations (e.g. micrognathia) and upper airway abnormalities (e.g. glossoptosis). A third cause, which seldom

occurs in association with the previous ones, is the presence of neuromuscular diseases that lead to a lack of upper airway control mechanisms. For these reasons, the diagnostic approach and therapeutic planning should involve several specialists<sup>1-5</sup>. Peculiarities of SDB in pediatric age include higher frequency of “obstructive hypoventilation” instead of periodic and distinct apneic events; poor correlation between clinical picture severity, daytime symptomatology and objective picture; daytime hyperactivity rather than sleepiness; normal sleep architecture compared to the sleep fragmentation observed in adults; and obstructive apnea observed only in the rapid eye movement (REM) phase<sup>1-5</sup>. Comorbidities can be present although they are more common in the adult population<sup>6</sup>. The broad term SDB also encompasses non-obstructive causes of sleep-related breathing disorders, such as sleep-related hypoventilation disorders and central sleep apnea syndromes. For example, the term “obstructive SDB” may include a child who snores and has hyperactive behaviors during the day, but his or her polysomnography (PSG) does not show OSA. Guilleminault et al<sup>7</sup> first described the increased Upper Airway Resistance Syndrome (UARS), having observed in some children a higher respiratory effort due to an increase in upper airway resistance. This condition is not associated with apneic episodes or pathological desaturations but only with flow limitations and microarousals. The clinical consequences of this syndrome, due to an increased muscular effort demanding excessive energy, are evidenced by insufficient weight growth (due to a reduction of the growth hormone secretion in sleep phase four), poor school performance, and daytime irritability. It is important to remember that the diagnosis of UARS may not be detected via PSG because of the lack of apnea and periods of hypoxemia. Diagnosis of UARS is based rather on clinical suspicion deriving from the evidence of flow limitations and microarousals, as inferred from developmental and behavioral cues. In cases in which a surgical treatment is planned and the clinical picture tends toward a medium-severe form, a severity balance must be defined by PSG or cardiorespiratory monitoring: in fact, the risk profile of postoperative complications is determined by the degree of severity of the OSA and the presence of preoperative comorbidities. The degree of severity must be defined by PSG or cardiorespiratory monitoring because the specificity of the questionnaires is equal to 100 for generic diagnosis of SDB but not for OSA, thus pulse ox-

imetry may be a useful screening method<sup>8,9</sup>. In recent years, OSA patients have been observed to show elevated inflammatory protein levels (C-reactive protein, interleukin 10, interleukin 6, fibrinogen, leukotrienes) proportional to the degree of severity; this preliminary evidence, if confirmed, may represent a low-cost valid test<sup>10</sup>. The objective of this review article is to evaluate the role of the otorhinolaryngologist in pediatric OSA.

## Materials and Methods

A literature review has been performed on articles retrieved from PubMed and Scopus from the last 30 years on the following topics: peculiarities of sleep-disordered breathing in pediatric age; discrimination of sleep disorders; adenotonsillar hypertrophy; surgical techniques; adjuvant surgical procedures.

## Results

### *The Role of the Otorhinolaryngologist*

The otolaryngologic clinical examination includes the oropharyngoscopy, which aims at evaluating the tonsillar volume, the space occupied by the tongue and the residual respiratory space, and a fibroscopy with flexible optics to evaluate the degree of collapsibility on the retropalatal and retrolingual level and the consequent degree of obstruction. However, it is necessary to consider the difficulty in performing a dynamic endoscopic examination in children. This may be a sufficient reason, especially in cases of medium-severe grade, to plan a preoperative drug-induced sedation endoscopy (DISE), also known as sleep-endoscopy. It is important to remember that the presence of tonsillar hypertrophy plays a major role in OSA in otherwise healthy children. On the other hand, tonsillar hypertrophy is non-determinant when there is a neuromuscular deficit, obesity or age under two years. These factors are themselves causes of persistence of apnea after adenotonsillectomy. It is widely accepted that isolated adenoid hypertrophy is not sufficient to determine apnea, but it can cause worsening in children at risk<sup>11</sup>. In addition, the objective examination must always evaluate the possible presence of reduced weight growth, or obesity; hyperactivity, restlessness, learning difficulty; right ventricular hypertrophy,

pulmonary hypertension; allergy and/or asthma; and neuromuscular disorders, syndromic states, mucopolysaccharidosis. These conditions are to be considered predictive factors for postoperative complications. Surgical removal of the tonsils and adenoids is considered the first-line treatment for OSA in otherwise healthy children over two years of age with adenotonsillar hypertrophy, as recommended by the American Academy of Pediatrics, the American Academy of Otolaryngology-Head and Neck Surgery and Italian guidelines from the Otolaryngology and Pediatrics Societies. Literature data demonstrates, in mild forms of OSA, a complete normalization of PSG data and quality of life after adenotonsillectomy. In these patients, alternatives to in-laboratory PSG are possible, such as “nap” PSG, overnight continuous pulse oximetry, or audio and video monitoring. However, all of these tests have a low negative predictive value; a negative result is insufficient to exclude OSA<sup>12</sup>. The possible complications related to the surgical procedure itself overlap with those found for adenotonsillectomy surgery performed for other indications. Significant increase in anesthesiological complications in the induction phase and in the phase of awakening is related to the severity of OSA or the presence of predictive factors (under 2 years of age, underweight, obesity, allergy, asthma, cardiorespiratory impairment, apnea hypopnea index (AHI) > 10/h, SaO<sub>2</sub> nadir < 70). The percentage of minor postoperative respiratory complications (desaturation < 90%, increased respiratory effort, laryngospasm, asthmatic crisis) varies between 16 and 27%. In contrast, with the presence of severe OSA, neuromuscular disorders or craniofacial malformations 1-15% of children have severe respiratory complications (desaturation < 8.0%, pleural effusion, pneumothorax, pulmonary hypertensive crisis)<sup>13</sup>. In these patients the intervention must be performed in hospitals that have intensive pediatric care. The purpose of PSG in these children is to improve diagnostic accuracy in high-risk populations and define the severity of OSA to optimize perioperative and postoperative planning.

### ***Surgical Techniques***

A variety of instruments have been devised for the removal of tonsillar and adenoid tissue. Traditionally, the most common approach has been extracapsular (complete) tonsillectomy, but intracapsular (partial) tonsillectomy is increasingly used. Extracapsular tonsillectomy (also known as complete, total, or sub-capsular tonsillectomy)

consists of removal of the entire palatine tonsil and the surrounding fascia or capsule with either cold or hot techniques. For “cold” or sharp dissection, the mucosa is sharply excised from the tonsillar pillar with a scissor or scalpel, and blunt instruments are used to divide the tonsillar capsule from the underlying musculature. Hemostasis is then achieved with a combination of pressure, cautery, ligation, or hemostatic agents. For “hot” techniques, electro-surgical or thermal instruments are utilized to excise the tonsillar tissue. A variety of instruments and technologies have been developed for this purpose including lasers, diathermy, bipolar electro-surgical scissors or forceps, and bipolar radiofrequency ionic dissociation. Each of these approaches has advantages. Hot techniques allow for simultaneous cauterization of blood vessels and enable surgeons to perform the procedure, in most cases, quickly and with minimal blood loss. Cold dissection and radiofrequency techniques are associated in some studies with less pain and more rapid return to normal function. None of the more current clinical guidelines advocate for use of any one specific surgical technique for extracapsular tonsillectomy over others, and the decision of which technique to use is generally based on surgeon preference and training<sup>14</sup>. Intracapsular tonsillectomy (also known as subtotal or partial tonsillectomy, or “tonsillotomy”) is increasingly used for the treatment of obstructive SDB. In this technique, microdebriders, bipolar electro-surgical scissors, or radiofrequency ablation (including “coblation”) devices are used to debulk the obstructing portions of the tonsil parenchyma<sup>15</sup>. Some evidence suggests that the technique permits more rapid recovery compared with traditional tonsillectomy, and possibly reduces the risk of postoperative hemorrhage. However, there is a risk of tonsillar regrowth, which usually is clinically insignificant but occasionally requires revisional surgery. Both extracapsular and intracapsular tonsillectomy are effective modalities to treat OSA, with comparable surgical outcomes on short-term follow-up. However, when comorbid diagnoses of both asthma and obesity exist, OSA is likely to be refractory to treatment with intracapsular compared with extracapsular tonsillectomy<sup>16</sup>. Adenoidectomy consists in the removal of the obstructing portions of adenoid tissue from the nasopharynx with the use of either sharp curettes (sharp or cold dissection), electro-cautery, coblation, or microdebrider. The procedure is generally done transorally after retraction

of the soft palate, taking care to avoid trauma to the undersurface of the palate or Eustachian tube orifices. Occasionally, tissue removal via a trans-nasal approach may also be necessary.

### **Adjuvant Surgical Procedures**

Surgical procedures other than adenotonsillectomy are sometimes considered to treat OSA in children without significant tonsillar hypertrophy, or with residual disease after adenotonsillectomy. These adjuvant surgical procedures may also be beneficial in patients with a high probability that OSA is due to factors other than adenotonsillar hypertrophy alone, such as in children with obesity, Down syndrome, craniofacial syndromes, or neuromuscular disease. They include tongue base procedures and expansion pharyngoplasty and lateral pharyngoplasty.

Airway obstruction at the level of the base of the tongue is increasingly recognized as a cause of persistent OSA after adenotonsillectomy, especially in children with obesity, Down syndrome, and craniofacial or neuromuscular disorders<sup>17</sup>. Endoscopic-assisted coblation lingual tonsillectomy and robotic surgery are the most used techniques for a volumetric reduction of the tongue base.

In some children, OSA is associated with collapse of the lateral pharyngeal wall. Surgical procedures to correct this problem include expansion sphincter pharyngoplasty or lateral pharyngoplasty.

## **Discussion**

The otorhinolaryngologist has a central role in the diagnosis and treatment of pediatric OSA. The diagnostic role is essential for the evaluation of the upper airways and of biometric and polysomnographic data, while the treatment can rely on medical therapies or surgical approach.

In the majority of healthy children, adenotonsillectomy for OSA results in a dramatic improvement in respiratory parameters as measured by polysomnography. There is moderately good evidence that adenotonsillectomy improves quality of life in children with OSA<sup>18</sup> and there is somewhat weaker evidence that adenotonsillectomy may improve cognitive function and behavior in children with OSA. Observational studies have reported postoperative improvements in objective measures of impulsivity, inattention, and cognitive function after adenotonsillectomy<sup>19</sup>.

DISE is used for the definition and management of patients with residual OSA after adenotonsillectomy. Even though the first work published in the literature on sleep endoscopy by Croft et al<sup>20</sup> in 1990 is on children, even today DISE has not found a complete, precise, and commonly accepted codification in patients of pediatric age<sup>21</sup>, both as regards the anesthetic protocol and in which cases to carry it out. Regarding the drugs to be used, since some of the children do not tolerate venous access, the initial use of inhalation anesthetics (halothane or sevoflurane) is a confusing factor for pediatric DISE because it alters the pharyngeal muscle tone.

The drugs used in DISE in children are propofol, dexmedetomidine (not approved by the Food and Drug Administration for pediatric use in the United States), ketamine with glycopyrrolate to reduce secretions and tachycardia, remifentanyl and midazolam.

DISE evaluates the extent and orientation of obstruction at four to five anatomical sites of potential collapse (adenoids, palate or velum, lateral pharyngeal wall, tongue base, and epiglottis or supraglottis)<sup>22</sup>.

A recent systematic review of the literature examined the scoring systems used to report the results of DISE in both pediatric and adult age. Among the 21 staging methods, the most used is Velum oropharynx tongue base epiglottis (VOTE), followed by the Pringle and Croft Classification<sup>23</sup>. Vicini in 2012 published a classification termed “Nose Oropharynx Hypopharynx and Larynx” (NOHL), using a system similar to the TNM staging for neoplasms. This classification, respecting the objectives of the DISE, takes into consideration the obstructive site and the degree and pattern of obstruction<sup>24</sup>. A systematic review on the use of pediatric DISE in 2016<sup>25</sup> concludes that DISE does not modify decisions in cases of significant adenotonsillar hypertrophy but should be used in cases where there is persistence of apnea after adenotonsillectomy. However, many children have residual OSA following adenotonsillectomy, as determined through PSG. Identification of a biomarker of residual disease would be clinically meaningful to detect children at risk. Bhattacharjee et al<sup>26</sup> have shown that the serum high-sensitivity C-reactive protein is an inflammatory biomarker, predictive of residual OSA after adenotonsillectomy. Resolution of OSA after adenotonsillectomy (post-AT) was defined by a post-AT AHI < 1.5/h. The residual OSA was defined as an AHI post-AT > 5/h, which is considered to be clinically significant.

## Conclusions

Otolaryngologic assessment is of paramount importance to correctly classify a child with OSA. Correct inspection of the upper airway and quantification of the quality of sleep through polysomnography lead to the right therapeutic choice. Knowledge of different surgical techniques helps to deal with residual OSA after studying the obstruction sites by DISE.

## Sources of Funding

This work was supported by the Italian Society of Rhinology. The sponsor provided financial support for costs related to the publication of this article. The sponsor was not involved in the study design, in the collection and interpretation of data, in the writing of the study, or in the decision to submit the article for publication.

## Conflict of Interests

The authors declare that they have no conflict of interest.

## References

- 1) COHEN-LEVY J, POTENZA J, COULOIGNER V. Pediatric obstructive sleep apnea syndrome: treatment strategy. *Arch Pediatr* 2017; 24: S39-S47.
- 2) ALVES RS, RESENDE MB, SKOMRO RP, SOUZA FJ, REED UC. Sleep and neuromuscular disorders in children. *Sleep Med Rev* 2009; 13: 133-148.
- 3) FREGOSI RF, QUAN SF, KAEMINGK KL, MORGAN WJ, GOODWIN JL, CABRERA R, GMITRO A. Sleep-disordered breathing, pharyngeal size and soft tissue anatomy in children. *J Appl Physiol* 2003; 95: 2030-2038.
- 4) LAM YY, CHAN EY, NG DK, CHAN CH, CHEUNG JM, LEUNG SY, CHOW PY, KWOK KL. The correlation among obesity, apnea-hypopnea index, and tonsil size in children. *Chest* 2006; 130: 1751-1756.
- 5) GOLDBERG S, SHATZ A, PICARD E, WEXLER I, SCHWARTZ S, SWED E, ZILBER L, KEREM E. Endoscopic findings in children with obstructive sleep apnea: effects of age and hypotonia. *Pediatr Pulmonol* 2005; 40: 205-210.
- 6) PAPACHATZAKIS I, VELENTZA L, ZAROGOLIDIS P, KALLIANOS A, TRAKADA G. Comorbidities in coexisting chronic obstructive pulmonary disease and obstructive sleep apnea - overlap syndrome. *Eur Rev Med Pharmacol Sci* 2018; 22: 4325-4331.
- 7) GUILLEMINAULT C, KHRAMTSOV A. Upper airway resistance syndrome in children: a clinical review. *Semin Pediatr Neurol* 2001; 8: 207-215.
- 8) BRIETZKE SE, KATZ ES, ROBERSON DW. Can history and physical examination reliably diagnose pediatric obstructive sleep apnea/hypopnea syndrome? A systematic review of the literature. *Otolaryngol Head Neck Surg* 2004; 131: 827-832.
- 9) STATHAM MM, ELLURU RG, BUNCHER R, KALRA M. Adenotonsillectomy for obstructive sleep apnea syndrome in young children: prevalence of pulmonary complications. *Arch Otolaryngol Head Neck Surg* 2006; 132: 476-480.
- 10) GOLDBART AD, TAL A. Inflammation and sleep disordered breathing in children: a state-of-the-art review. *Pediatr Pulmonol* 2008; 43: 1151-1160.
- 11) KAY DJ, MEHTA V, GOLDSMITH AJ. Perioperative adenotonsillectomy management in children: current practices. *Laryngoscope* 2003; 113: 592-597.
- 12) MARCUS CL, BROOKS LJ, DRAPER KA, GOZAL D, HALBOWER AC, JONES J, SCHECHTER MS, WARD SD, SHELDON SH, SHIFFMAN RN, LEHMANN C, SPRUYT K, AMERICAN ACADEMY OF P. Diagnosis and management of childhood obstructive sleep apnea syndrome. *Pediatrics* 2012; 130: e714-755.
- 13) SACCHETTI ML, DI MASCIO MT, TINELLI E, MAINERO C, RUSSO G, FIORELLI M, CALISTRI V, DE LENA C, MINNI A, CARAMIA F. Resting state functional thalamic connectivity abnormalities in patients with post-stroke sleep apnoea: a pilot case-control study. *Eur Rev Med Pharmacol Sci* 2017; 21: 2676-2689.
- 14) PINDER DK, WILSON H, HILTON MP. Dissection versus diathermy for tonsillectomy. *Cochrane Database Syst Rev* 2011: CD002211.
- 15) FRIEDMAN M, WILSON MN, FRIEDMAN J, JOSEPH NJ, LIN HC, CHANG HW. Intracapsular coblation tonsillectomy and adenoidectomy for the treatment of pediatric obstructive sleep apnea/hypopnea syndrome. *Otolaryngol Head Neck Surg* 2009; 140: 358-362.
- 16) MUKHATIYAR P, NANDALIKE K, COHEN HW, SIN S, GANGAR M, BENT JP, ARENS R. Intracapsular and extracapsular tonsillectomy and adenoidectomy in pediatric obstructive sleep apnea. *JAMA Otolaryngol Head Neck Surg* 2016; 142: 25-31.
- 17) WOOTTEN 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: 983-987.
- 18) VENEKAMP RP, HEARNE BJ, CHANDRASEKHARAN D, BLACKSHAW H, LIM J, SCHILDER AG. Tonsillectomy or adenotonsillectomy versus non-surgical management for obstructive sleep-disordered breathing in children. *Cochrane Database Syst Rev* 2015: CD011165.
- 19) AVIOR G, FISHMAN G, LEOR A, SIVAN Y, KAYSAR N, DEROWE A. The effect of tonsillectomy and adenoidectomy on inattention and impulsivity as measured by the Test of Variables of Attention (TOVA) in children with obstructive sleep apnea syndrome. *Otolaryngol Head Neck Surg* 2004; 131: 367-371.
- 20) CROFT CB, PRINGLE M. Sleep nasendoscopy: a technique of assessment in snoring and obstructive sleep apnoea. *Clin Otolaryngol Allied Sci* 1991; 16: 504-509.
- 21) LOVATO A, KOTECHA B, VIANELLO A, GIACOMELLI L, STAFFIERI A, MARCHESE-RAGONA R. Nasal and oral snoring endoscopy: novel and promising diagnostic tools in OSAS patients. *Eur Arch Otorhinolaryngol* 2015; 272: 1793-1799.
- 22) CHAN DK, LIMING BJ, HORN DL, PARIKH SR. A new scoring system for upper airway pediatric sleep endoscopy. *JAMA Otolaryngol Head Neck Surg* 2014; 140: 595-602.

- 23) DURR ML, MEYER AK, KEZIRIAN EJ, ROSBE KW. Drug-induced sleep endoscopy in persistent pediatric sleep-disordered breathing after adenotonsillectomy. *Arch Otolaryngol Head Neck Surg* 2012; 138: 638-643.
- 24) VICINI C, DE VITO A, BENAZZO M, FRASSINETI S, CAMPANINI A, FRASCONI P, MIRA E. The nose oropharynx hypopharynx and larynx (NOHL) classification: a new system of diagnostic standardized examination for OSAHS patients. *Eur Arch Otorhinolaryngol* 2012; 269: 1297-1300.
- 25) GALLUZZI F, PIGNATARO L, GAINI RM, GARAVELLO W. Drug induced sleep endoscopy in the decision-making process of children with obstructive sleep apnea. *Sleep Med* 2015; 16: 331-335.
- 26) BHATTACHARJEE R, KHEIRANDISH-GOZAL L, KADITIS AG, VERHULST SL, GOZAL D. C-reactive protein as a potential biomarker of residual obstructive sleep apnea following adenotonsillectomy in children. *Sleep* 2016; 39: 283-291.