

Obstructive sleep apnea syndrome in the pediatric age: the role of the dentist

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Abstract. – OBJECTIVE: Sleep disordered breathing in children designates a wide spectrum of respiratory disorders characterized by partial or complete obstruction of the upper airways. It ranges from primary snoring, its mildest clinical manifestation, to obstructive sleep apnea syndrome (OSAS): complete obstruction of the upper airways with cessation of airflow. The aim of this paper is to highlight the roles of the pediatric dentist and the orthodontist in the therapeutic approach to pediatric OSAS as a “sentinel” who can detect early signs of the disease for immediate referral to the otolaryngologist and as an active participant in therapy.

MATERIALS AND METHODS: A literature review has been performed on the following topics: pediatric OSAS, orthodontic clinical aspects of pediatric OSAS, orthodontic therapy of pediatric OSAS, mandibular advancement devices and functional orthodontic devices in OSAS treatment.

RESULTS: The role of the dentist in pediatric OSAS is essential to correct orthodontic alterations that may favor the development of the condition. Orthodontic treatment aims at reducing the severity of OSAS by increasing the airspace and improving airflow through orthopedic expansion of the upper jaw and mandibular advancement. Rapid palatal expanders and mandibular advancement devices are successfully used in the treatment of OSAS.

CONCLUSIONS: Scientific evidence of a strong association between craniofacial growth and OSAS; the pediatric dentist and the orthodontist participate as sentinels, observing and identifying conditions requiring referral to the otolaryngologist and playing a pivotal role in the orthodontic treatment phase.

Key Words

OSAS, Pediatric age, Dentist, Sleep disordered breathing.

List of Abbreviations

SDB: sleep disordered breathing; PS: primary snoring; OSAS: obstructive sleep apnea syndrome; MADs: mandibular advancement devices.

Introduction

Sleep disordered breathing (SDB) is commonplace in children. It is caused by neurophysiological changes occurring during sleep due to variations in the muscle tone of the pharyngeal walls; these variations in tone, in association with anatomic variations, are most commonly induced by tonsillar and adenoidal hyperplasia, leading to breath disorders during sleep^{1,2}.

SDB causes in children can be grouped by age. In small children, airway flow may be reduced due to nasal obstruction, to impaired skeletal anatomy, to impaired soft tissues, or to other neuromuscular causes. On the other hand, childhood obesity has been identified in recent studies as a major cause of obstruction in older children³.

Successful treatment of SDB in children depends on accurate identification of the site of obstruction and assessment of its severity. SDB in children exhibits a severity spectrum ranging from primary snoring (PS), its mildest clinical manifestation, to obstructive sleep apnea syndrome (OSAS), its most severe form^{4,5}. While primary snoring is not associated with blood-gas exchange anomalies, or sleep fragmentation or deconstruction, OSAS is characterized by partial or complete obstruction of the upper airway that impairs normal ventilation during sleep. Airflow reduction, known as hypopnea, or cessation, known as apnea, during sleep are associated with the shrinkage of the pharyngeal space down to its total collapse. OSAS in children is characterized by apnea episodes lasting more than 5 seconds, in association with an up to 4% oxyhemoglobin reduction, hypercapnia, arousal, and persistence of thoracic and/or abdominal respiratory movements⁵.

The OSAS pathogenic mechanism establishes itself in conditions of hypoventilation linked to physiological hypotonia of the pharyngeal muscles during sleep, associated with a pathological

reduction of oropharyngeal space, with retro-position of the tongue, increased nasal resistance, and shrinking of the upper airways during sleep⁶.

The dentist and the orthodontist play a central role in pediatric OSAS. The aim of this paper is to highlight the roles of the pediatric dentist and the orthodontist in the therapeutic approach to pediatric OSAS both as a “sentinel” who can detect early signs of the disease for immediate referral to the otolaryngologist and as an active participant in therapy.

Materials and methods

A literature review has been performed on articles retrieved from PubMed and Scopus from the last 40 years on the following topics: pediatric OSAS, orthodontic clinical aspects of pediatric OSAS, orthodontic therapy of pediatric OSAS, mandibular advancement devices and functional orthodontic devices in OSAS treatment.

Results

Historical Background

In 1889, William Hill recognized the symptoms of OSAS in children as those of a real illness, while in 1892, William Osler noticed an association between adenotonsillar hypertrophy and sleep disordered breathing. In 1918, Osler noted similarities between obese and hypersomnial patients and a character described by Charles Dickens in “The Posthumous Papers of the Pickwick Club.” In 1956, Burwell et al⁷ defined the Pickwickian syndrome as one that included obesity, hypercapnia, pulmonary heart, erythrocytosis, daytime hypersomnolence, and respiratory disturbances. In 1976, Guilleminault et al⁸ first defined OSAS as a distinct clinical entity characterized by clinical signs and evidence of obstructive apnea. The first polysomnography dates back to 1976 by Guilleminault himself and to date it is confirmed to be the gold standard for OSAS diagnosis³.

Prevalence

Pediatric OSAS prevalence varies between 1% and 3%³, while most of authors report a prevalence of 10% among children presenting with primary snoring. The etiology of pediatric OSAS recognizes adenotonsillar hypertrophy as the principal cause of upper airway obstruction⁹. This creates a maximum incidence peak between 2 and 8 years,

when adenotonsillar tissue occupies a larger volume in comparison to available airway space.

Favoring Conditions in the Pediatric Age Range

The main etiology of pediatric OSAS recognizes adenoidal and tonsillar hypertrophy as its primary cause; however, several additional risk factors should also be considered. Predisposing conditions for increased nasal resistance, and other factors, may play a decisive role in occurrence of the syndrome: anomalies of the maxillary structure, such as micrognathism and retrognathism, or of the soft tissues, such as macroglossia, favor a reduction of the oropharyngeal space^{5,10,11}. Obstructive nasal diseases, such as allergic rhinitis, are also pediatric determinants of nasal congestion to take into consideration¹². Other predisposing factors include age, obesity, familiarity, cranio-facial dysmorphism, and syndromic pathologies such as Down syndrome or Pierre Robin sequence³.

The Role of the Dentist

The effects of breathing alterations in general and of airway obstructions in particular on craniofacial growth continue to be an amply discussed topic in pediatric medical and dental literature¹³.

Most studies investigating correlations between pathologies of the respiratory tract and craniofacial alterations in the growing subject showed that a chronic and protracted mouth-breathing pattern during childhood corresponds to postural abnormalities of both skeletal and soft tissues that identify a specific cephalometric stereotype¹³⁻¹⁶. This is defined by an increased mandibular post-rotation, an increased lower front facial height, an increased inclination of the mandibular plane, a reduced ratio of the posterior facial height to the total frontal height, and an increased maxillo-mandibular discrepancy on the sagittal plane^{14,17,18}. This stereotype induces in these children a vertical growth with dolichofacial typology. Recent studies^{17,19} show that the cephalometric stereotype associated with respiratory dysfunction varies according to the obstructive tissue involved. In particular, Baroni et al¹⁹ highlights that the long face stereotype corresponds predominantly to an adenoidal hypertrophy, while tonsillar hypertrophy is mainly characterized by a tendency to a more horizontal mandibular growth, a tendency of the jaw to rotate counterclockwise, and a higher ratio between the posterior and anterior facial heights. A stereotype similar to the dolichofacial growth pattern was found in subjects with SDB. Many authors study-

ing the association between SDB and alterations in craniofacial growth highlighted a dolichofacial growth pattern, an increased lower frontal facial height, post-rotation of the mandibular plane, contraction of the upper jaw, high and narrow palate, and maxillary and mandibular crowding^{5,9,20}.

However, in the pediatric context OSAS patients must be split into two classes: mouth breathing and non-mouth breathing. In the first case, craniofacial growth abnormalities are those described above and include a dolichofacial growth pattern, an increased lower frontal facial height, a post-rotation of the mandibular plane, and a concomitant reduction in the inter-maxillary space and in the pharyngeal air space. The posture of the tongue, low and protruded with a contracted upper arch, is also pathognomonic. In these patients, the dentist has the task of identifying the clinical features of an adenoid facies, with labial incompetence associated with intraoral orthodontic features such as contracted upper jaw, crossbite, and increased mandibular angle²⁰.

The craniofacial characteristics of non-mouth breathing OSAS subjects are different. In this case, a deep bite with type II skeletal class and mandibular retraction (Figure 1) is common. This retraction, coupled to a high and retracted position of the tongue, favors the pathogenic mechanism of OSAS. In these subjects, the tongue posture is not associated with a contracted superior arch with ogival palate, as in the first group. Rather, the arch morphology will present a wider maxillary cross-section. In the non-mouth breathing OSAS patients, the dentist should detect the presence of a deep upper maxilla with small and retracted jaw⁹.

The orthodontist's clinical evaluation of subjects affected by OSAS is, therefore, based on the recognition of an adenoid facies, micrognathia, retrognathia, and ogival palate. Table I summarizes the main orthodontic issues and the therapeutic goals of the two classical OSAS phenotypes here described.

Finally, as part of the clinical examination, the orthodontist will have to distinguish the classical OSAS phenotype, corresponding to children with adenotonsillar hypertrophy, with or without skeletal malocclusion, and the congenital OSAS phenotype, where anomalies such as micrognathia or craniofacial alterations are associated with genetic syndromes, e.g. Pierre Robin sequence.

Orthodontic Therapy of Pediatric OSAS

Orthodontic treatment aims at reducing the severity of OSAS via orthopedic expansion of the upper jaw and/or mandibular advancement, thus increasing the airspace and improving airflow. Polysomnographic clinical evidence after orthodontic treatment suggests that orthodontic therapy improves airway patency. The rapid palatal expander appliance (Figure 2) increases nasal volume, stimulates the growth of the jaw complex and improves nasal ventilation in the OSAS child.

A recent systematic review of the literature points out that the success of orthodontic treatment of pediatric OSAS is linked to the expansion of airways⁹. Mandibular advancement devices (MADs), which facilitate the lowering of the mandible, also reduce the collapsibility of the airway and stimulate the airway dilator muscles



Figure 1. Deep bite in a non-mouth breather pediatric subject with obstructive sleep apnea syndrome (OSAS).

Table I. Orthodontic and therapeutic aspects of pediatric OSAS.

Orthodontic issue	Therapy goal
High and retracted tongue	Lingual repositioning
Class II	Class correction
Deep bite	Bite correction
Mandibular retrusion	Mandibular advancement
Hypodivergence	Growth control

The main orthodontic issues and the therapeutic goals of the two classical OSAS phenotypes in children.

(Figure 3). Evidence shows that MADs reduce the values of apnea/hypopnea indices, though they do not completely normalize them. In addition, current studies on the use of MADs in the treatment of pediatric OSAS show that the recorded improvements of OSAS-related parameters are short-term.

An action similar to MADs can be obtained with functional orthodontic devices used in the treatment of class II patients with deep bite²¹. Among these, the recent adoption of preformed plastic devices improved comfort for children affected by OSAS, thus favoring adherence to treatment (Figure 4). These devices are inserted in the frontal part of the oral cavity and have niches for housing the dental elements of both arches. Their action induces an advancement of the bite. The advantage of these devices is the ability to combine orthodontic therapy with myofunctional therapy aimed at restoring intra- and extraoral muscle balance and restoring normal nasal ventilation^{21,22}.

Discussion

SDB's ample clinical variability has a strong impact on a child's quality of life; effects may include reduced statural development, morning headaches, enuresis, attention disorders with effects on school activities, daytime sleepiness, on up to cardiorespiratory complications^{1,4,6}.

Poor recognition of pediatric SDB in clinical practice is related to the fact that 80% of symptomatic habitual snorers are not reported to the pediatrician⁵. As primary snoring is the mildest level of SDB, if not recognized it might progressively lead to an increase in airway resistance, up to the occurrence of OSAS. An early diagnosis of SDB in children is, therefore, crucial. Since the observed symptoms present a wide clinical variability, a multidisciplinary approach to the pathology becomes indispensable and must involve the pediatrician, the otorhinolaryngologist, the orthodontist, the neurologist, the nutritionist, and the physiotherapist³.

Snoring is the main symptom of OSAS. Hence, the importance of a pediatric visit is always investigating sleep habits and the presence of night snoring. The basic diagnosis is referred to the otolaryngologist, but the orthodontist can intervene in correcting occlusal problems associated with OSAS.

Orthodontic treatment aims at reducing the severity of OSAS via orthopedic expansion of the upper jaw and/or mandibular advancement, thus increasing the airspace and improving airflow. These approaches, regardless of the specific selection suited to the patient's characteristics and therapeutic



Figure 2. Rapid palatal expander in a mouth breather pediatric subject with obstructive sleep apnea syndrome (OSAS).

Figure 3. Mandibular advancement device (MAD).



needs, reduce the child's nighttime apnea episodes, as confirmed by polysomnographic exams on treated pediatric patients. Orthodontic therapy thus can and should be integrated into a multidisciplinary approach to treating the child with OSAS²³.

In March 2016, the Italian Health Ministry published national guidelines for the prevention and treatment of OSAS in the pediatric age range. The pediatric OSAS diagnostic pathway begins with validated questionnaires administered to parents, goes through a clinical examination, and must be confirmed by instrumental examination. During the clinical examination, the dentist must pay attention to craniofacial features such as elongated face, small and retracted jaw, deep bite, contracted upper jaw, and deep palate. The dentist, in the case of confirmed snoring or OSAS in a growing patient and in the presence of related craniofacial morphology, may apply fixed rapid maxillary

expansion devices and/or mandibular propulsion devices, as appropriate.

Patients with OSAS-related craniofacial and occlusal morphology, coupled with a history of snoring, inability to breathe with the nose, allergies, asthma, and/or obesity, should be sent to the pediatrician for a multidisciplinary evaluation and subsequently to the otorhinolaryngologist for further instrumental examinations, among which polysomnography represents the gold standard to determine the presence and severity of OSAS²⁴.

Conclusions

Scientific evidence of a strong association between craniofacial growth and sleep breathing disorders such as OSAS suggests a multidisciplinary approach to the management of the problem. In this



Figure 4. Preformed functional device.

approach, the professional figures of the pediatric dentist and the orthodontist participate as sentinels, observing and identifying conditions requiring referral to the otolaryngologist. In addition, they play a pivotal role in the orthodontic treatment phase that, through orthopedic expansion of the upper jaw or mandibular advancement, contributes to ameliorating the severity of childhood OSAS.

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Conflict of Interests

The authors declare that they have no conflict of interest.

References

- 1) BROUILLETTE RT, FERNBACH SK, HUNT CE. Obstructive sleep apnea in infants and children. *J Pediatr* 1982; 100: 31-40.
- 2) LUZZI V, DI CARLO G, SACCUCCI M, IERARDO G, GUGLIELMO E, FABBRIZI M, ZICARI AM, DUSE M, OCCASI F, CONTI G, LEONARDI E, POLIMENI A. Craniofacial morphology and airflow in children with primary snoring. *Eur Rev Med Pharmacol Sci* 2016; 20: 3965-3971.
- 3) LI HY, LEE LA. Sleep-disordered breathing in children. *Chang Gung Med J* 2009; 32: 247-257.
- 4) CARROLL JL, MCCOLLEY SA, MARCUS CL, CURTIS S, LOUGHLIN GM. Inability of clinical history to distinguish primary snoring from obstructive sleep apnea syndrome in children. *Chest* 1995; 108: 610-618.
- 5) KATYAL V, PAMULA Y, DAYNES CN, MARTIN J, DREYER CW, KENNEDY D, SAMPSON WJ. Craniofacial and upper airway morphology in pediatric sleep-disordered breathing and changes in quality of life with rapid maxillary expansion. *Am J Orthod Dentofacial Orthop* 2013; 144: 860-871.
- 6) ROSEN CL. Obstructive sleep apnea syndrome (OSAS) in children: diagnostic challenges. *Sleep* 1996; 19: S274-277.
- 7) BURWELL CS, ROBIN ED, WHALEY RD, BICKELMANN AG. Extreme obesity associated with alveolar hypoventilation--a Pickwickian Syndrome. 1956. *Obes Res* 1994; 2: 390-397.
- 8) GUILLEMINAULT C, ELDRIDGE FL, SIMMONS FB, DEMENT WC. Sleep apnea in eight children. *Pediatrics* 1976; 58: 23-30.
- 9) NAZARALI N, ALTALIBI M, NAZARALI S, MAJOR MP, FLORES-MIR C, MAJOR PW. Mandibular advancement appliances for the treatment of paediatric obstructive sleep apnea: a systematic review. *Eur J Orthod* 2015; 37: 618-626.
- 10) PAHKALA R, PUUSTINEN R, TUOMILEHTO H, AHLBERG J, SEPPA J. Risk factors for sleep-disordered breathing: the role of craniofacial structure. *Acta Odontol Scand* 2011; 69: 137-143.
- 11) COSTA ESRA, DOS SANTOS GIL NA. Craniofacial skeletal architecture and obstructive sleep apnoea syndrome severity. *J Craniomaxillofac Surg* 2013; 41: 740-746.
- 12) LUZZI V, IERARDO G, VISCOGLIOSI A, FABBRIZI M, CONSOLI G, VOZZA I, VESTRI A, POLIMENI A. Allergic rhinitis as a possible risk factor for malocclusion: a case-control study in children. *Int J Paediatr Dent* 2013; 23: 274-278.
- 13) BASHEER B, HEGDE KS, BHAT SS, UMAR D, BAROUDI K. Influence of mouth breathing on the dentofacial growth of children: a cephalometric study. *J Int Oral Health* 2014; 6: 50-55.
- 14) LESSA FC, ENOKI C, FERES MF, VALERA FC, LIMA WT, MATSUMOTO MA. Breathing mode influence in craniofacial development. *Braz J Otorhinolaryngol* 2005; 71: 156-160.
- 15) SOUSA JB, ANSELMO-LIMA WT, VALERA FC, GALLEGO AJ, MATSUMOTO MA. Cephalometric assessment of the mandibular growth pattern in mouth-breathing children. *Int J Pediatr Otorhinolaryngol* 2005; 69: 311-317.
- 16) OSIATUMA VI, OTUYEMI OD, KOLAWOLE KA, OGUNBANJO BO, AMUSA YB. Occlusal characteristics of children with hypertrophied adenoids in Nigeria. *Int Orthod* 2015; 13: 26-42.
- 17) FRANCO LP, SOUKI BQ, CHEIB PL, ABRAO M, PEREIRA TB, BECKER HM, PINTO JA. Are distinct etiologies of upper airway obstruction in mouth-breathing children associated with different cephalometric patterns? *Int J Pediatr Otorhinolaryngol* 2015; 79: 223-228.
- 18) DREVENSEK M, PAPIĆ JS. The influence of the respiration disturbances on the growth and development of the orofacial complex. *Coll Antropol* 2005; 29: 221-225.
- 19) BARONI M, BALLANTI F, FRANCHI L, COZZA P. Craniofacial features of subjects with adenoid, tonsillar, or adenotonsillar hypertrophy. *Prog Orthod* 2011; 12: 38-44.
- 20) ALTALIBI M, SALTAJI H, RODUTA ROBERTS M, MAJOR MP, MACLEAN J, MAJOR PW. Developing an index for the orthodontic treatment need in paediatric patients with obstructive sleep apnoea: a protocol for a novel communication tool between physicians and orthodontists. *BMJ Open* 2014; 4: e005680.
- 21) COZZA P, POLIMENI A, BALLANTI F. A modified monobloc for the treatment of obstructive sleep apnoea in paediatric patients. *Eur J Orthod* 2004; 26: 523-530.
- 22) GASPARINI G, AZZUNI C, RINALDO FM, CERVELLI D, MARIANETTI TM, SFERRAZZA A, PELO S. OSAS treatment with oral appliance: assessment of our experience through the use of a new device. *Eur Rev Med Pharmacol Sci* 2013; 17: 385-391.
- 23) ROSE E, RIDDER GJ, STAATS R. Endoscopically-assisted adjustment of an oral appliance in patients with obstructive sleep apnoea. *Laryngorhinootologie* 2002; 81: 619-623.
- 24) IERARDO G, LUZZI V, POLIMENI A. Obstructive Sleep Apnea Syndrome (OSAS): evaluation and treatment of odontostomatological problems. *Med Lav* 2017; 108: 293-296.