Sleep Disorders in Pediatric Dentistry

Clinical Guide on Diagnosis and Management

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Preface

Our world has been always in a constant change; things we used to do only 10 or 20 years ago could be obsolete, proven wrong, or replaced with new ideas. This applies to so many things, and the medical and dental world is no exception into this. This fast pace of change has implication to any profession, and without any doubt, every dentist has experienced this in his or her career.

Dentistry as it was taught 20 or more years ago was more conventional; it was considered as a separate discipline that only borders medicine. Dentistry is still taught as a profession for people with extreme good manual dexterity; most dentist can remember the dental school admission requirement of sculpting a soap bar. More and more emphasis is given to other qualities like:

- A desire to learn: Having a willingness to learn new things and a desire to improve your skills is a great trait. This is the best way to cope with constant change.
- Good problem-solving skills: Problem-solving skills are an essential trait for a dentist. Not every patient will have a dental/medical problem with a clear-cut solution. Sometimes a dentist needs to think outside the box in order to determine the best treatment approach for the patient.

  Becoming a dentist takes an interest in science and a desire to help people.

Dentistry slowly becomes an important partner in healthcare; we have seen publications of correlations of the common gingivitis and heart disease [1]. Oral healthcare professionals can identify patients who are unaware of their risk of developing serious complications as a result of cardiovascular disease and who are in need of medical intervention.

We have seen publications about the value of early detection of oral cancer [2]. The prevention of oral cancer and its associated morbidity and mortality hinges upon the early detection of neoplastic lesions.

The oral cavity is an important anatomical location with a role in many critical physiologic processes, such as digestion, respiration, and speech. The mouth is frequently involved in conditions that affect the skin or other multi-organ diseases. In many instances, oral involvement precedes the appearance of other symptoms or lesions at other locations [3].

The latest trend is the involvement of dentistry in the management of obstructive sleep apnea [4]. The general dentist can play an important role in the recognition of signs and symptoms of patients with obstructive sleep apnea syndrome. Obstructive

sleep apnea (OSA) is defined as the repetitive airway obstruction due to the collapse of the pharyngeal airway during sleep potentially causing partial or complete cessation of airflow for breathing. Although many suffer from symptoms of sleep apnea syndrome, most remain undiagnosed until significant problems occur, such as cardiopulmonary disease, neurologic dysfunction, and hypersomnolence. In recent years, sleep apnea has become a significant public health concern. Both medical and dental practitioners have become increasingly aware of sleep apnea. Early detection of this condition by the dental practitioner can lead to the prevention of comorbid diseases and improved quality of life for many patients.

General dentists are involved in the management of OSA of adults by providing alternative treatment with oral appliances (common acronyms: OAT, oral appliance therapy; MAD, mandibular advancement device; MAS, mandibular advancement splint). The gold standard for the treatment of OSA (adults and children) has been, and still is, the use of positive airway pressure (PAP) treatment. The immediate treatment outcome of the use of a PAP device is usually excellent; however, compliance is an issue. Adherence to positive airway pressure treatment for obstructive sleep apnea is a critical problem with adherence rates ranging from 30% to 60%. Poor adherence to PAP is widely recognized as a significant limiting factor in treating OSA, reducing the overall effectiveness of the treatment and leaving many OSA patients at heightened risk for comorbid conditions, impaired function, and quality of life [5]. Dentists who have received additional training can provide alternative treatment with OAT, and commonly the adherence rate is better than with PAP therapy [6].

OSA does not only affect adults; it does also affect children. The health consequences of OSA on adults are well documented [7]. There is now a wealth of information indicating that untreated obstructive sleep apnea is associated with an increased risk of fatal and nonfatal cardiovascular event, a higher propensity of sudden death during sleep, and a greater risk for stroke and all-cause mortality.

The health consequences for children with OSA are even more compelling. We have begun to understand that pediatric sleep disorders in general, and more particularly sleep-disordered breathing (SDB), can lead to substantial morbidities affecting the central nervous system (CNS), the cardiovascular and metabolic systems, and somatic growth, ultimately leading to reduced quality of current and future life. We have also monitored the altered behavior of children with OSA [8]. This can be confusing and can sometimes lead to a wrong diagnosis and treatment. Behavioral problems are prevalent in children with either mild SDB or OSA, and both groups of children show significant improvements in behavior after treatment. Research has also identified a link between sleep disorders and problematic and hyperactive behaviors and mood disturbances [9].

Charles Dickens has in 1836 described in his *The Pickwick Papers* a boy, called Joe, that has the characteristic of a person who suffers from sleep-disordered breathing [10]. Unknowingly, he described a child with pediatric SDB. This was the first description in the main literature about sleep-disordered breathing; for that reason, SDB is also initially known as the “Pickwickian syndrome.”

The process of understanding pediatric sleep apnea is ongoing, and I am sure there is still a lot to be discovered. We already understand that pediatric sleep apnea has other dimensions than adult sleep apnea. Children are not just small people; strategies that apply to adults do not automatically apply to children. Side effects of OSA (adult) treatment are sometimes magnified when applied to children. As an example, we see that the compression forces of the mask of a PAP device limit the craniofacial growth of the child, potentially making the craniofacial structure more unfavorable for a normal (nasal) breathing.

There was a pilot study [11] with the pediatric application of MAD (oral appliances) that was designed for adults; after the initial promising results, we have not seen any further publications. One can only guess why this study has been terminated.

The most common cause of pediatric OSA is the enlargement of lymphoid tissue: adenoids and lingual tonsils. The enlargement of these tissues can create a structural blockage of the upper airway and higher airflow resistance. The most common treatment is the surgical removal of these tissues. Not enough attention is given why in the first place these tissues are hypertrophied; alternative management and understanding of these tissues will be researched in the coming years. Most importantly is that we need to develop a strategy to avoid adenotonsillar hypertrophy in the first place.

Quite commonly, children that are diagnosed with OSA do have a dysfunction in the way they breathe, speak, eat, and drink. Quite often, the children have an impaired nasal breathing leading to mouth breathing. A dysfunctional orofacial system is one of the causes of disturbed craniofacial growth and teeth crowding. A correction or, even better, reeducation of these functions has been shown in the literature as an effective (co-)treatment of pediatric OSA [12]. Current literature demonstrates that myofunctional therapy decreases apnea-hypopnea index by approximately 50% in adults and 62% in children! Myofunctional therapy could serve as an adjunct to other obstructive sleep apnea treatments.

The most promising treatment option of treatment of pediatric OSA is currently dentofacial orthopedic and orthodontics. This option has the ability to correct structural issues to improve nasal and pharyngeal airways. This allows to normalize structures so the orofacial complex can function how they should function. There are many publications [13–15] that report reduction of the apnea-hypopnea index after the expansion of the palate through rapid palatal expansion (RPE). It is believed that palatal expansion increases the palatal tongue space and nasal airway and facilitates nasal breathing.

In cases where there is a midfacial deficiency, a combination of palatal expansion with the use of a reverse pull headgear (protraction headgear) seems to have very good results [16].

One of the unknown and underestimated facts is that dentistry is in the best position to recognize the signs and symptoms of pediatric sleep apnea; dentists see children from a young age (2 years and up) on a scheduled regular base; they do inspect the orofacial region, and they could detect or recognize any signs or

symptoms that could be related to pediatric sleep disorders. Dentists will also be able to observe the behavior and mood; this could sometimes be a sign of a pediatric sleep disorder.

However, at this time, there is a lack of awareness and education about this very important topic, and this book is one of the contributions to correct that void.

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It is impossible to list everybody who has influenced me on my journey, so I will name the 4 most influential persons in my professional career as a dentist:

- Dr. John Witzig whose passion sparks me to explore the world of functional orthodontics
- Dr. Brock Rondeau who gave me a solid foundation in orthodontics
- Dr. John Mew who gave me the understanding of the importance of "oral posture"
- Dr. Steven Olmos who taught me the connection of TMD and OSA

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Last but not least, I thank all the coauthors of this book; without their willingness to share knowledge and their hard work, this book would never have seen the daylight.

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Obstructive Sleep Apnea Syndrome

1

Manisha Budhdeo Witmans

1.1 Introduction

Sleep is a basic physiological need, and humans spend about one third of their lives sleeping. Sleep architecture is composed of two basic types of sleep called rapid eye movement (REM) sleep and non-REM (NREM) sleep. NREM sleep can be further subdivided into stages N1, N2, and N3, which make up different portions of the night that the individual spends sleeping. The amount of each stage can change on the basis of the person's age. Less is known about gender differences in sleep. The different sleep stages are characterized by different electroencephalogram patterns and electro-oculography patterns. NREM sleep makes up 75% of the night (5% is stage N1 sleep, 45–55% is stage N2 sleep, and about 25% is stage N3 sleep), while REM sleep accounts for the remaining 25% [1]. In contrast to children and adults, infants spend almost 50% of the time in REM sleep, and as much as 80% of sleep is spent in REM sleep in premature infants. Physiologically, humans are most vulnerable to perturbations in breathing during REM sleep, thus often classified as sleep-related breathing disorders when there are associated disturbances in gas exchange. The primary sleep disorder associated with breathing abnormalities in REM sleep is obstructive sleep apnea (OSA). Different names are used to describe the spectrum of this disorder and may include upper airway resistance syndrome, sleep apnea, obstructive apnea, sleep-disordered breathing, and obstructive sleep apnea hypopnea syndrome. This form of sleep-disordered breathing is different from central sleep apnea and hypoventilation, which are related to other pathophysiological mechanisms and involve different treatment options. The reader is encouraged to read the *International Classification of Sleep Disorders* (3rd Edition) for more comprehensive discussion and explanations about the various sleep-related breathing disorders. The focus of the following discussion will be relevant to obstructive sleep apnea. This is arguably a very important disorder in sleep medicine, as it has many

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serious consequences for the affected individuals, including increased morbidity and mortality and for society in general as its impact affects performance, vigilance, and optimal functioning. Unfortunately, at this time, although there are treatment options that may help manage the resulting symptoms and prevent complications, we are certainly nowhere near the point of lifelong cure. The treatment may alleviate or reduce symptoms and sequelae for a period of time; age is a risk factor for persistence or recurrence of the disorder. However, we remain hopeful that advances in science and technology will improve the management of sleep apnea and enable us to develop more customized and individual specific treatment options.

1.2 History

Obstructive sleep apnea was initially reported in the 1970s. It has become increasingly prevalent and is a significant public health problem. Descriptions of OSA in the lay literature can be traced back to Shakespeare, as well as the famous character Joe from *The Pickwick Papers*, the Charles Dickens novel. Use of the term Pickwickian syndrome is discouraged because this term is not only used to describe obstructive sleep apnea but also has been used to describe individuals with obesity hypoventilation syndrome. In the medical literature, Dr. John Cheyne was the first person to describe sleep-disordered breathing associated with heart failure and an irregular breathing pattern during sleep [2]. Cases were slowly reported over time until the mid-twentieth century, when the problem became widely recognized, and its consequences extend to every sphere of medicine. Obesity was thought to be the primary factor in the development of sleep apnea, until the 1970s, when Drs. Dement and Guilleminault showed that sleep apnea could occur in thin individuals [1]. They were instrumental in establishing the field of sleep medicine and first described OSA in children in 1976 [1]. Since then, the field has expanded exponentially to become a subspecialized field of medicine, rooted in several disciplines, including respirology, neurology, psychiatry, psychology, pediatrics, otolaryngology, internal medicine, cardiology, anesthesia, and dentistry.

Obstructive sleep apnea is a disorder characterized by recurrent episodes of partial upper airway obstruction (hypopnea) or complete upper airway obstruction (apnea) during sleep, despite respiratory efforts, and it results in sleep disruption, usually an arousal, and ventilatory instability [3] (drops in oxygen saturation, and swings in blood pressure during the apneic episodes). The Task Force of the *International Classification of Sleep Disorders* (3rd Edition) has defined obstructive sleep apnea in children and adults separately, particularly because of the differences in the diagnostic criteria noted below. The spectrum of disordered breathing during sleep can range from snoring to frank OSA with its associated consequences. Snoring is more prevalent in children and adults compared to OSA, with rate estimates ranging from 17% to 33% in men versus 7–19% in women (Principles and Practice of Sleep Medicine). OSA associated with daytime sleepiness affects 3–7% of adult men and 2–5% of adult women [1]. Depending on the

criterion used, the lowest estimates suggest 4% prevalence in males and 2% prevalence in females. A recent systematic review determined the prevalence rates to be widely variable in adults, based on the threshold for defining sleep apnea, and estimated rates as high as 49%! In some older age groups, the estimates were higher than 80% [4]. In children the prevalence rates of OSA vary from 1% to 5%, depending on the diagnostic criteria used to define OSA. The disease defining quantitative parameter used to calculate the frequency and severity of airflow obstruction is called the apnea–hypopnea index (AHI) (Table 1.1), measured during overnight polysomnography (PSG). The American Academy of Sleep Medicine (AASM) has classified the severity of sleep apnea on the basis of cutoffs for apnea/hypopnea. Mild, moderate, and severe OSA are classified as ≥5, ≥15, and ≥30 events per hour, respectively. OSA-related symptoms include excessive daytime sleepiness, morning headaches, behavioral mood problems, insomnia, and identified comorbidities such as hypertension. The challenge of this definition is that using this as the only indicator of disease is that it fails to provide information regarding the physiological and/or functional impact of this disorder on affected individuals. Various candidate genes such as TNFa have been linked to phenotypes of OSA and are being evaluated.

What is not understood with absolute certainty is the threshold of change from benign snoring to OSA along the continuum of breathing during sleep. Snoring may certainly be disruptive to a bed partner or the affected individual but does not result in any reportable consequences. In contrast, it is arguable that any snoring reflects airflow limitation in the airway, and any resulting consequences may not be appreciated until the severity of the problem is significant enough to affect the bed partner or to result in sleepiness, daytime impairment, and/or cardiovascular consequences.

Table 1.1 Types of events associated with obstructive sleep apnea, according to the American Academy of Sleep Medicine (*AASM*) [5] scoring manual in adults. In children, the same definitions apply but the duration is for greater than or equal to 2 breaths and not 10 seconds. Children 13–18 can be scored using adult criteria.

Obstructive apnea: Decrease in the peak airflow signal excursion by 90% of the pre-event baseline for 10 s with continued or increased respiratory effort throughout the entire period of absent airflow
Hypopnea: Decrease in the peak airflow signal excursion by 30% of the pre-event baseline for 10 s with associated arousal or 3% oxygen desaturation with any one of the following: snoring, increased inspiratory flattening of the airflow signal, or associated thoracoabdominal paradox
Central apnea: Decrease in the peak airflow signal excursion by 90% of the pre-event baseline for 10 s with absent respiratory effort throughout the entire period of absent airflow
Central hypopnea: Decrease in the peak airflow signal excursion by 30% of the pre-event baseline for 10 s with associated arousal or 3% oxygen desaturation with none of the following: snoring, increased inspiratory flattening of the airflow signal, or associated thoracoabdominal paradox
Hypoventilation: Increase in the arterial PCO_2 to a value 55 mmHg for 10 min or an increase in arterial PCO_2 by 10 mmHg from the awake baseline to a value >50 mmHg for 10 min. In children, this is defined as hypercapnia, or increased $PCO_2 > 50$ mmHg for at least 25% of the total sleep time.

PCO_2 partial pressure of carbon dioxide

In fact, snoring alone has been associated with excessive daytime sleepiness, and those who snore tend to have a higher Epworth sleepiness score (ESS) >10 [6]. In children, snoring has been associated with poorer executive function. As in other disorders, many of the origins of adult sleep apnea may stem from infancy or childhood. Children with OSA have been found to have elevated blood pressure 10–15 mmHg higher than nonsnoring controls during sleep, irrespective of the severity of the sleep apnea [7]. OSA is a highly prevalent and serious chronic disease with significant morbidity and mortality and with increasing prevalence worldwide [8]. If it does indeed start in childhood, this behooves us to address the problem early and comprehensively.

1.3 Pathogenesis

Obstructive sleep apnea occurs because there is a lack of adequate compensation to maintain an open airway when the normal reduction in pharyngeal dilator muscle activity is superimposed on a narrowed airway with increased collapsibility [9]. A Starling resistor model is used to explain the human pharyngeal airway during sleep [10]. However, the inspiratory flow decreases with increasing effort which is called negative effort dependence, rather than being fixed as predicted by the Starling model. Wellman and colleagues have shown that the resistance in the upper airway can vary considerably in patients with sleep apnea and in turn influence downstream narrowing as well [11]. Although the pathogenesis of OSA has not been conclusively determined, certain factors have been identified that are attributable to obstructive sleep apnea. (1) Pharyngeal anatomy and collapsibility determine the critical closing pressure, which is the pressure at which the airway will narrow or close as described above and its inherent variability within individuals. This has been shown to be less negative, meaning more collapsible, in children and adults with sleep apnea. (2) Ventilatory control system gain or loop gain, which is the responsivity of the system to deal with perturbations in respiratory control. Therefore, a high loop gain will promote apneas as a response to the initial disturbance because of overcompensation, whereas a low loop gain will reduce the perturbations in breathing [12]. (3) The ability of the airway to dilate or stiffen in response to an increase in ventilatory drive. (4) Arousal threshold is the point at which an individual may respond to the apnea and the associated perturbations with a cortical arousal to an apnea. (5) Fluid shift which refers to the increased volume of venous fluid and/or upper airway mucosal fluid may contribute to the decreased surface area and increased resistance during sleep, which may decrease the volume of the airway during sleep [13]. Elegant work by Marcus and colleagues in children have shown that children with obstructive sleep apnea have impaired two-point discrimination in the tongue and palate compared to healthy controls [14]. They also showed that the palate is not affected as it is in adults when using vibration perception as a measurement in small number of control with sleep apnea [15]. Taken together,