Rapid Maxillary Expansion and Adenotonsillectomy in 9-Year-Old Twins With Pediatric Obstructive Sleep Apnea Syndrome: An Interdisciplinary Effort

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Pediatric obstructive sleep apnea is known to cause neurocognitive problems, yet it often goes undetected or mistreated. The authors describe 9-year-old twins with snoring, enlarged tonsils, and excessive daytime sleepiness whose symptoms had been previously disregarded by health care professionals. At presentation, a dentist found the patients to be midface deficient and symptomatic. A home sleep test, prescribed by the dentist, revealed apnea-hypopnea index readings of 74/h and 16/h, respectively. The children were referred to an otolaryngologist, and a continuous positive airway pressure therapy trial resulted in improved cognition and temperament. Rapid maxillary expansion was then performed at the dentist office, followed by adenotonsillectomy by an ear, nose, and throat specialist and myofunctional rehabilitation with a speech pathologist for both patients. After treatment, results mimicked those reported during the continuous positive airway pressure trial, with substantially reduced apnea-hypopnea index of 0.9/h and 1.6/h. This case highlights the interdisciplinary nature of pediatric obstructive sleep apnea management and the need for all health care professionals to receive comprehensive sleep medicine training for proper diagnosis and treatment.

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Sleep-disordered breathing (SDB) is the umbrella term used for nighttime breathing disorders that range from primary snoring without symptoms to obstructive sleep apnea (OSA). The prevalence of SDB in the general pediatric population ranges from 7% to 11%,1 and approximately 90% of children with SDB have their condition go undiagnosed.2 These rates are concerning, as SDB has been associated with irreversible neurocognitive problems in children.3,4 According to Jan et al.,5 “…untreated chronic sleep disorders may lead to impaired brain development, neuronal damage and permanent loss of developmental potentials.” As a result, children with undiagnosed SDB have been reported to use health care 226% more than the general population.6

Sleep-disordered breathing is a multifactorial disease and requires a multidisciplinary team—including osteopathic, allopathic, ear, nose, and throat (ENT) specialist, dental, speech, and myofunctional professionals—to manage the condition appropriately. Pediatric SDB symptoms likely present frequently in the general patient population of each of these different fields.
For example, an infant presenting to a pediatrician’s office with feeding or latching problems may have lingual ankylosis (ie, “tongue tie”), and a child presenting to a myofunctional or speech pathologist clinic with interdentalized speech sounds (s,z,t,d,n,l) may have tongue thrust. Left unmanaged, these conditions may manifest in teenagers as midface deficiency and class II malocclusion requiring orthodontic treatment or behavioral issues requiring psychiatric care. Therefore, it is important that all health care professionals are able to recognize SDB symptoms.

A myriad of reasons can contribute to airway restriction in children with SDB, including genetics and fetal development. According to Lee et al, pediatric sleep apnea in nonobese children is a disorder of craniofacial growth. Children who cannot breathe well through their nose will breathe through their mouth. Orthodontic researchers Harvold et al described how oral breathing changes the functional relationship established by the sum of all the soft tissues operating with the craniofacial bones; the functional soft-tissue matrix is the epigenetic governing determinant of the skeletal growth process. Thus, oral breathing is an orofacial dysfunction that may lead to dysmorphism and the onset of pediatric OSA.

Enlarged tonsils and adenoids are often responsible for nasal obstruction in the pediatric population, forcing patients to mouth breathe. Adenotonsillectomy is the first line of treatment for patients with OSA. However, this procedure may not be enough to resolve OSA, particularly if the patient’s underlying craniofacial disorder is severe. Rapid maxillary expansion (RME), an orthodontic procedure used to manage structural and functional problems in the midface, consists of application of orthopedic forces to the midpalatal suture, with the forces dissipating across the cranial and circum-maxillary sutures.

Specifically, RME widens the maxilla and the nasal cavity base; this increase in nasal volume decreases airway resistance and improves respiratory patterns. With improved nasomaxillary structures, patients with oral myofunctional disorders must be rehabilitated through myofunctional re-education.

We describe the case of 9-year-old female twins who presented to a dental office with symptoms of severe OSA that were previously dismissed by 2 health care professionals. This case demonstrates the lack of knowledge about SDB among pediatric health care professionals and the importance of an interdisciplinary approach to treatment.

Report of Case

Nine-year-old female twins (patients A and B) presented to a dental office with loud snoring, excessive daytime sleepiness, and enlarged tonsils. Their mother reported that they had been irritable and moody on awakening each morning. The twins had previously been examined by their pediatrician and an ENT specialist, both of whom had not recommended further testing or treatment. Visual clinical assessment revealed both patients to be midface deficient with a crowded oropharyngeal space. A standard pediatric sleep questionnaire was administered by the dentist. Both patients scored 12 out of 22 possible points, with a score of 8 or higher indicating need for a sleep evaluation. The mother also presented a video of patient A sleeping, which showed snoring and apnea.

A home sleep study (HST) was ordered, which was performed with a 4-channel (ie, effort, flow, pulse, and O2 saturation) device. The HST findings revealed an apnea-hypopnea index (AHI) of 74/h for patient A and 16/h for patient B.

Severe OSA was diagnosed by the sleep physician, and a treatment plan consisting of rapid maxillary expansion (RME) with the dentist for 7 weeks, followed by an adenotonsillectomy by an ENT specialist was initiated. After 2 months of recovery, myofunctional rehabilitation was administered for 3 months with the speech pathologist. A trial of continuous positive airway pressure (CPAP) at 7 cm H2O was conducted for both patients to establish a baseline for treatment results. With the CPAP, AHI for both patients dropped to 0/h, and after the first night, the mother reported dramatic improvement in cognition and temperament. Although CPAP is
common therapy for adult patients with OSA, it is not a long-term solution for pediatric patients because their facial skeleton is still developing. When CPAP is used for a prolonged period in this patient population, it can restrict growth of the facial skeleton and cause structural changes that may exacerbate the issue.

Figure 1. Home sleep study findings of 9-year-old twins (patients A and B) with pediatric obstructive sleep apnea before and after treatment (rapid maxillary and adenotonsillectomy). Normalization of the breathing is visually seen in the decreased amplitude of the waveforms.
Both patients continued temporary CPAP therapy during RME, for a total of 7 weeks. After RME and the cessation of CPAP therapy, HSTs revealed AHIs of 11/h for patient A and 4/h for patient B. The AHIs further improved after adenotonsillectomy, with scores of 0.9/h for patient A and 1.6/h for patient B. Findings for HSTs conducted before, during, and after treatment are shown in Figure 1. For the

Figure 1. Continued.
After treatment, positive subjective results from both patients mimicked those observed during the CPAP trial. The mother and patients reported a substantial increase in quality of life, with both patients exhibiting

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Figure 1. Continued.
improvements in energy levels, ability to focus, social skills, and overall happiness. At the 12-month follow-up, AHIs were 1.1 for patient A and 1.0 for patient B (Figure 2).

### Discussion
Interdisciplinary health care has received an increasing amount of attention over the past decade. In numerous reports, the Institute of Medicine has proposed a future
in which collaboration and interdisciplinary team-based care are standard in health care delivery.18,19 This concept is now being applied in various health care environments, as well as within health care professionals’ education and training.20 However, to our knowledge, few reports have objectively demonstrated the beneficial effects of an interdisciplinary approach to pediatric OSA management in a private clinic setting initiated by a dental professional.

The present case is unique in that its large collection of data (Figure 1 and Figure 2) showed improvement on HSTs after each stage of treatment. These objective findings illustrate how collaboration and treatment planning by health care professionals from 3 distinct fields of medicine (ie, dental, ENT, and myofunctional rehabilitation) were necessary to address the various issues presented by the patients’ severe OSA. As previously mentioned, SDB is a multifactorial disease affecting several different musculoskeletal structures and functions; thus, members of the multidisciplinary team should be chosen according to individual patient needs. For example, pediatric psychologists may address SDB-related behavioral issues, whereas osteopathic physicians may use osteopathic manipulative treatment to address postural or pain issues that result from the forward head posture that is common in patients with SDB.21-23 Additional research is warranted to measure the effectiveness of such multidisciplinary treatment approaches.

High-functioning multidisciplinary treatment teams understand that SDB is dynamic and persistent, with symptoms often recurring after treatment.11 As pointed out by Guilleminault and Sullivan,24 the ultimate goal of SDB is the restoration of continuous nasal breathing. If mouth breathing recurs at any point after treatment, the patient may experience abnormal airway growth or compromised neuromuscular response in airway tissues. Therefore, treatment teams must recognize and monitor any underlying structures, functions, or behaviors impairing optimal breathing. Of note, the present patients’ concerns were dismissed by 2 health care professionals, despite the severity of the condition. This experience is in agreement with previous findings that pediatricians and other primary care physicians are often unprepared to manage sleep disorders.25,26 According to Owens,25 despite empirical evidence, inadequate attention is often paid by health care professionals to sleep disorders and their serious health consequences. This lack of attention may be related to the fact that sleep and sleep disorders traditionally receive little attention in medical schools’ curricula.26

Polysomnography (PSG)—the preferred method for diagnosing pediatric OSA27—was never ordered in the present case. By the time the patients and their mother presented to the dental office, they chose to undergo HSTs rather than return to their pediatrician for a PSG referral. This situation is not uncommon—in our
experience, some patients may not have access to or wish to subject their children to an overnight stay in a laboratory, which can be costly and traumatic. Research indicates that home sleep apnea testing may be a viable alternative to PSG in such cases.\textsuperscript{28} However, health care professionals should pursue PSG before HST because of HSTs’ poor negative predictive value.\textsuperscript{29} The limitations of the HST should be considered when interpreting the findings in the present case.\textsuperscript{15}

Another factor contributing to low rates of pediatric OSA diagnosis may be the limitations in current screening methods. Pediatric sleep questionnaires such as the one used in the present case\textsuperscript{15} are an invaluable screening tool for SDB, but they lack a basic tool of differential diagnosis: clinical observation of craniofacial structures. As previously mentioned, pediatric SDB in the nonobese patient is a craniofacial dysmorphism similar to class II orthodontic malocclusion.\textsuperscript{30} Changes in the craniofacial hard tissue are often recognizable in the esthetics of the patient’s face. In the present case, the dentist was able to see physical signs of SDB by performing a head and neck examination, which in turn prompted him to administer the questionnaire.

Research\textsuperscript{30} has suggested that clinical assessment of craniofacial features considered as risk factors for SDB can be helpful. Even a simple assessment of the patient’s facial profile can help clue the provider. Educating physicians to screen by evaluating patients’ jaw and other facial structures may be an important interdisciplinary opportunity to improve pediatric SDB screening methods.

Conclusion

Severe pediatric OSA can often be overlooked by health care professionals. The present case demonstrates how an interdisciplinary treatment approach can yield dramatic improvement in sleep study findings and quality of life. Management of pediatric OSA is an interdisciplinary effort in which physicians, dentists, and myofunctional therapists are key members. It is essential for health care professionals from all disciplines to receive proper sleep medicine training to ensure early diagnosis and treatment.

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References


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