

Towards Restoration of Continuous Nasal Breathing as the Ultimate Treatment Goal in Pediatric Obstructive Sleep Apnea

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Abstract

The interaction between oral-facial structural growth and muscle activity starts early in development and continues through childhood. Chronic oral breathing is an important clinical marker of orofacial muscle dysfunction, which may be associated with palatal growth restriction, nasal obstruction, and/or a primary disorder of muscular or connective tissue dysfunction. It is easily documented objectively during sleep.

Treatment of pediatric obstructive-sleep-apnea (OSA) and sleep-disordered-breathing (SDB) means restoration of continuous nasal breathing during wakefulness and sleep; if nasal breathing is not restored, despite short-term improvements after adenotonsillectomy (T&A), continued use of the oral breathing route may be associated with abnormal impacts on airway growth and possibly blunted neuromuscular responsiveness of airway tissues.

Elimination of oral breathing, i.e., restoration of nasal breathing during wake and sleep, may be the only valid end point when treating OSA. Preventive measures in at-risk groups, such as premature infants, and usage of myofunctional therapy as part of the treatment of OSA are proposed to be important approaches to treat appropriately SDB and its multiple co-morbidities.

Keywords

Obstructive sleep apnea; Pediatrics; Oral-facial muscles; Nasal-oral functions; Myofunctional-therapy

Pediatric sleep disordered breathing (SDB) is seen in children along the entire spectrum of body mass index (BMI). Whether underweight, overweight, or within normal BMI limits, when pediatric SDB is present, there is abnormal collapsibility of the upper airway leading to abnormal breathing during sleep. This abnormal breathing is commonly associated with snoring, and severity of the upper airway collapse has been polygraphically defined. Importantly, such definitions rely in part on the type of recording performed during sleep, the sophistication of the sensors used to investigate abnormal breathing, and the experience of the interpreter; abnormalities from primary snoring and "nasal flow limitation" to complete "obstructive sleep apnea" (OSA) have been defined [1-3].

Remarkably, studies have shown that independent of the type of abnormal breathing during sleep noted on polysomnography (PSG), negative daytime consequences of pediatric SDB have been observed that may be sub-classified as neuro-behavioral, cardiovascular, and/or inflammatory [4-6]. However, not all children with abnormal sleep suffer from each of these consequences. Considering the above comorbidities, phenotyping of pediatric SDB patients has been suggested, with an overweight/obese subgroup of children, who may tend to have more nocturnal desaturation, and possibly cardiovascular and metabolic comorbidities; and a normal weight subgroup of children who may tend to present with neurobehavioral complaints, including problems with focus and concentration, hyperactivity, non-REM sleep parasomnias, learning problems, headache, mood disturbances, for example [7-12].

First Line Treatment with Adenotonsillectomy and Decreased in Benefits Over Time

Whether overweight or not, often the tonsils and adenoids in children with SDB are found to be enlarged. Clinically, it is recognized that enlargement of these tissues is often associated with frequent oral breathing. Historically, adenotonsillectomy (T&A) has been performed to treat obstructive sleep apnea in children since the late 1970s [13], though it was certainly used before this in the setting of enlarged tonsils and adenoids. T&A came to be considered the first-line of therapy for pediatric SDB, and earlier studies, usually with short follow-up durations and with a variety of endpoints, suggested that T&A was highly successful for SDB in children, in both normal weight and overweight subgroups.

Over the years, systematic follow-up studies have revealed that T&A may not be as successful as once thought. [14-21] For years, routine post-operative PSG recordings were deemed unnecessary and expensive; so when subjective clinical report of improvement was observed post operatively, no further PSG testing was performed. However, increasingly there have been reports indicating that T&A may not be a reliable cure for SDB [16-21]. Of particular interest is how SDB fares in children in the long run, whether treated or not. Most research on natural history of treated and/or untreated pediatric OSA are biased towards the short-term, towards relatively younger children, and may not involve objective PSG measures. Reports on very long term follow-up (for example, 3 years or greater) of children with SDB, after T&A or not, are still rare, but suggest that sleep disordered breathing (AHI or symptoms) cannot be expected to remain resolved or significantly improved in the longer run [22-25]. The reports that do exist typically involve children who have presented again due to appearance of further clinical SDB-related symptoms, with OSA detected upon repeat testing. Intriguingly, snoring itself, even without hypoxia and frequent arousals, is associated with day time cognitive and behavioral morbidity similar to that seen with more pronounced nocturnal breathing abnormalities, which may suggest the importance of anatomy in the long run. One early article to attract attention focused on the orofacial structures as the predisposing factor involved in SDB "recurrence" [14]; however, until recently the substantial body of knowledge regarding the continuous interaction between normal breathing, particularly during sleep, and normal orofacial growth was not integrated into the sleep medicine field, despite longstanding and accepted understanding of such mechanisms in the dental and orthodontic fields [26-29].

A solid understanding of the factors that influence normal growth of the upper airway is critical to providing appropriate, long-sighted treatment to children with SDB. Based on important recent findings, it appears that complete SDB treatment may mean normalization of nasal breathing during sleep. Unfortunately, this outcome - continuous nasal breathing during sleep - is almost always ignored in pediatric PSG interpretation, even though the data is collected and available to analyze.

At this time, we are aware of only one study has reported systematic clinical, psychometric, and PSG follow-up evaluation of prepubertal children with SDB, who were enrolled at baseline and followed prospectively. This Taiwanese study involved 2 groups of children aged 6 to 12 years; and 4 to 6 years [25]. After T&A for SDB, follow-up occurred over 3 years, with systematic evaluation at 6, 12, 24, and 36 months post-surgically. Results at each time point were compared to pre-surgery findings. Independent of age group, this study demonstrated retention of about 70% of the initial group.

There was substantial improvement of symptoms and PSG findings at 6 months post-T&A, with about 50% of children having a normal apnea-hypopnea index (AHI). However, a progressive recurrence of clinical complaints and reemergence of abnormal PSG findings during the following 2.5 occurred, affecting both incompletely resolved SDB at 6 months, as well as those children with normal test results at 6 months post T&A. About 25% of the children with normal PSG results at 6 months still normal findings at the end of the study. (Bonuck and colleagues found in a large, longitudinal study of symptoms associated with SDB that adenoidectomy lowered the risk of future SDB symptoms by about 40-50%). [24] An interesting finding in the Taiwanese study came from comparison of the 2 age groups over time: the younger group had less "recurrence", and when recurrence was present, it took longer to reappear and was less severe. The investigators concluded that: 1) It is important to recognize the SDB syndrome early; 2) It is important to perform T&A at an early age if SDB is present; and 3) even with early intervention, a large portion of children with SDB will redevelop SDB overtime.

We propose that one reason for high rates of re-emergence of SDB in susceptible children is that normal nasal breathing has not been completely or lastingly reestablished after T&A, contributing to facial growth alterations and/or orofacial muscle tone deficits that predispose to further SDB over time. The importance of adequate nasal airway development and patency, the absence of which is clinically seen as mouth breathing, is suggested by both experimental findings and in a variety of clinical scenarios, described below.

Mouth breathing is common [24] - reported in 10-25% of children [30] - but as a marker or contributor to sleep disordered breathing, its role is largely unstudied. Intriguing associations exist, and are provided in detail below.

Interactions Between Orofacial Function and Growth: Experimental Data Involving Nasal Obstruction

The observation that increased nasal resistance and its companion, chronic oral breathing, alter facial growth is by no means new in medicine - Meyer described "adenoidal facies" in 1868, in which nasal obstruction from adenoidal hypertrophy led to what he termed "long face syndrome". Other have also commented on the apparent relationship between function and form [28]-i.e., obstruction and "deviant facial growth."

The craniofacial growth consequences of frequent mouth breathing may predispose to SDB. Mouth breathing has been demonstrated to lead to changes in muscle recruitment in the upper airway, which then alter craniofacial growth [27,31]. Small studies have evaluated the influence of oral breathing due to nasal obstruction on dento-facial development [32-34]. Over thirty years ago, a series of experiments in which nasal obstruction was induced in Rhesus monkeys for the first six months of life demonstrated that blockage of the nasal passages led to narrowing of dental arches, decreased maxillary arch length, and increased anterior facial height, as well as anterior cross-bite and maxillary overjet. [35-37] In these studies, EMG activity of oral facial muscles, including the geniohyoid and genioglossal muscles of the tongue, the suprahyoid dorsal tongue fibers, the upper lip elevators, and the digastric muscles, was shown to be abnormal in the monkeys with nasal obstruction. These experiments related morphometric skeletal changes to changes in muscle tone, which were present in the setting of continuous mouth breathing.

In humans, abnormal masseteric contractions have also been demonstrated in the presence of mouth breathing [38], suggesting that abnormal orofacial muscle activity links nasal obstruction to deficits in structural airway growth. Secondary posture changes associated with chronic mouth breathing have also been identified [30,39,40]. Interestingly, in the Rhesus monkey model, removal of nasal obstruction at 6 months led to return of normal nasal breathing and yielded improved morphometric development, whereas continued impairment of normal nasal breathing led to continued mouth breathing and abnormal oral-facial growth and development.

Interactions Between Orofacial Function and Growth: Observations in Disorders Involving Upper Airway Muscle Dysfunction

Increased nasal resistance is unlikely to be the sole avenue to chronic oral breathing and subsequent craniofacial growth alterations. In humans, neuromuscular disorders provide further insight about the relationship between altered muscle tone and changes in craniofacial development. [41-43] for example, in the myotonic dystrophies and some congenital myopathies, abnormal orofacial muscle tone leads to impaired development of craniofacial structures. Presentation includes increased vertical facial growth, a narrower maxillary arch, and deeper palatal depths. In these disorders, abnormal orofacial muscle tone has consequences for the growth of upper airway structures, in association with early and chronic mouth breathing and frequent development of obstructive SDB, with rates reported to be 43-69%.

Ehlers-Danlos Syndrome (EDS), on the other hand, is an inherited connective tissue disorder involving abnormal collagen. The collagen-vascular mutations seen in Ehlers-Danlos syndrome lead to abnormal facial growth. These changes lead to narrow nasal passages, forcing mouth breathing, particularly during sleep [44]. Clinical evaluation demonstrates abnormally long facial shape, narrow and/or high maxillary hard palate, often with crossbite. While initially only abnormalities of the naso-maxillary complex may be seen, as patients get older, defects of the mandibular condyle may become evident, which we hypothesize is promoted by the presence of chronic oral breathing. A similar pattern of facial growth abnormality is noted with dental agenesis: Mutations in homeobox genes including those involved in normal tooth development (including those with ectodysplasin A -EDA- and WNT 10A genes as noted in our patients) lead to narrow facial skeleton, mouth breathing and, in our study, to SDB [45-48].

History of prematurity is another circumstance associated with higher likelihood of sleep disordered breathing in childhood, and is therefore another interesting example of the interplay between muscle tone, craniofacial growth, and nasal versus oral breathing route. Recently a large convenience cohort of 300 premature infants (36 to 27 weeks gestational age) was followed for 3 years after birth with clinical evaluation, psychometric testing, facial and oral dimension assessment, and PSGs at birth, 12, 24 and 36 months of age. [49-50] as expected, the infants had a variable degree of hypotonia, with severity generally related to degree of prematurity. High and narrow hard palate (HNP) was noted at birth in many premature infants and was more common with younger gestational age; HNP infants were more likely to exhibit mouth breathing; and their mean apnea-hypopnea index (AHI) was significantly higher compared to the non-high/narrow palate group; and the HNP infants were also found to have significantly more feeding difficulties. While many infants with feeding difficulties did not receive early feeding/orofacial education services, including sensory stimulation training and oral-facial exercises, 42 infants did receive these services and rather remarkably, demonstrated improvements in palatal dimensions at 36 months relative to those without orofacial training. We hypothesize that orofacial muscle development played a role in normalization of palatal structures at 36 months.

There were also 23 infants who had a normal palate at birth, but evolved toward HNP, mouth breathing and SDB, suggesting that postnatal developmental factors also alter palatal growth [49].

In summary, whether experimentally induced or developmentally provoked, science and nature have provided with examples of the interplay between increased nasal resistance and/or poor muscle tone leading to chronic oral breathing, and subsequent altered craniofacial dimensions. We believe that the presence of chronic oral breathing is both a marker of an inadequate or obstructed nasal-pharyngeal airway, and a marker of persisting abnormalities in the developmental interplay between muscular control, breathing route, and structural growth of the upper airway.

Applications in the Treatment of Pediatric Sleep-Disordered Breathing

While the above considerations are suggestive, much more work is needed to understand chronic mouth breathing as a marker of, and possible precipitator of, SDB in pediatrics. To further understand the proposed detrimental role of abnormal orofacial tone and mouth breathing during sleep, PSGs of 64 non-obese children aged 3 to 9 years (with mean AHI=8.5 events/hour and mean flow limitation= 76%), and who had PSGs pre- and post- treatment for SDB, were assessed [51]. In our lab, an oral-only sensor (utilizing an oral scoop) is used to accurately and simply monitor mouth breathing [52]. In all of the baseline PSGs of the 64 children with SDB, there was evidence of excessive mouth breathing (defined as at least one third of total sleep time) on baseline diagnostic PSG. After adenotonsillectomy, 26 children had an AHI equal or higher than 1.5 events/hour. These children continued to have evidence of significant oral breathing. An additional 9 children whose AHI was under 1.5 events per hour also continued to have oral breathing - this is a very interesting group deserving further study. Clinically, children with SDB and persistent chronic mouth breathing after T&A may be referred for myofunctional therapy [53] in addition to usual therapies (e.g., consideration of anti-inflammatory medications, rapid maxillary expansion, CPAP). Eighteen children returned for 12 month follow-up, with only 9 having completed 6 months of myofunctional therapy. Though the numbers are very small, those who completed myofunctional therapy in addition to usual therapies were observed to have had improvements in nasal breathing as well as sleep, as measured by AHI and nasal flow limitation, beyond improvements seen in children without myofunctional therapy [51]. This suggests that even after nasal obstruction has been alleviated, improving muscle function of certain airway muscles, including the tongue, may improve function and/or growth of the upper airway, with resultant consequences for nasal breathing during sleep [51-55].

Observations and Conclusions: The Interplay Between Muscle Activity, Structural Growth, and Breathing During Wake and Sleep

The interaction between orofacial structural growth and muscle activity starts early in development, and the physiologic functions of suction, mastication, swallowing and nasal breathing in infancy play an important role in stimulating subsequent growth [55-58]. In the service of these functions, orofacial muscle use serves to help stimulate the direction and degree of growth. Mouth breathing is associated with altered oral-facial muscle activity and oral-facial growth. As such, its persistence is never normal. In fact, oral breathing has been termed "the most obvious manifestation of a syndromic pattern" involving a circuit of frequent infections, development of malocclusion, incorrect phonation, abnormalities of body posture, and changes in sleep. [30] Fortunately, oral breathing as a clinical sign has the advantage that its presence can be detected by simple direct observation, and its severity during sleep can be quantified with PSG.

During the past several decades, efforts have been undertaken to develop programs that will foster normal development of orofacial functions in at-risk children, including appliances as well as speech therapy, even, it could be argued, without recognition of all of the many benefits of doing so. Reeducation programs targeting normal orofacial muscle function have been developed in many countries, particularly among the orthodontic field, where oral-facial growth problems are often first identified [30,55,59-60]. Variants of myofunctional therapy have also been used in muscular dystrophies to delay secondary impacts on craniofacial bone growth and maxilla-mandibular impairment, and in young children to correct speech abnormalities, another common consequence of improper orofacial/ genioglossal tone, coordination, and/or structure. Despite these many applications, it is only in the recent past that myofunctional therapy has been proposed to make an impact in the treatment of SDB [30,53-54]. This is somewhat surprising, since muscle retraining has been used in adults with OSA with reduction of AHI, even without a proposed impact on the facial skeleton. Timing is likely to be important, since the gains from therapy are proposed to be via a mechanism of improved nasal breathing and improved craniofacial growth. Unfortunately, except for very limited reports, usage of myofunctional therapy very early in the course of SDB in pediatrics is limited, despite the fact that these therapies have existed for a long time. Thinking broadly, it could be argued that all of the accepted therapies for pediatric SDB may target improved nasal airflow one way or another; adding muscle strengthening might be an additional tool towards encouraging optimal craniofacial growth and perhaps long term improved outcomes in those at risk for SDB.

We conclude that oral breathing is an important clinical marker of orofacial muscle dysfunction, which may be associated with palatal growth restriction, nasal obstruction, and/or disorders of musculoskeletal dysfunction. Framing full treatment of pediatric SDB as restoration of continuous nasal breathing during wakefulness and sleep ought not to be considered. Our view based on the collected data is that if nasal breathing is not restored, despite short-term improvements after T&A, continued use of the oral breathing route will be associated with abnormal impacts on airway growth and possibly blunted neuromuscular responsiveness of airway tissues, both of which may predispose to the eventual return of upper airway collapse in later childhood, or in the full blown syndrome of OSA in adulthood. We believe elimination of oral breathing, i.e., restoration of nasal breathing during wake and sleep, may be the only valid "finish line" in pediatric sleep disordered breathing.

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