

## **Critical Review: Does orofacial myofunctional therapy improve outcomes for children with obstructive sleep apnea?**

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This critical review examines the evidence surrounding the benefits of orofacial myofunctional therapy in children with obstructive sleep apnea (OSA). A literature search resulted in four relevant articles, including a randomized control trial, two retrospective chart reviews, and a prospective case control study. Articles were appraised based on design, methods, analyses and degree of clinical relevance. Overall, all studies provided evidence that OMT should be considered as an adjunct treatment to an adenotonsillectomy, which is the current standard of care for most children with OSA.

### ***Introduction***

Obstructive sleep apnea (OSA) is a condition where part or all of the airway is blocked during sleep, resulting in multiple hypoxic events throughout the night (Kansagra & Vaughn, 2013). OSA is commonly diagnosed when the child has one or more hypoxic events an hour, combined with symptoms of sleep-disordered breathing (SDB) (Guilleminault et al., 2013; Villa et al., 2014; Villa, Evangelisti, Martella, Barreto & Del Pozzo, 2017). It is a frequently overlooked condition that is treatable. In a 2008 study, it was reported that the prevalence of OSA in children is approximately 1-4% (Lumen & Chervin, 2008), with obesity as an independent risk factor (CL et al., 2012).

Symptoms of pediatric obstructive sleep apnea include a reluctance to waking in the morning, increased irritability, decreased attention, headaches in the morning and behavior problems. These can lead to problems learning and decreased performance in school (Kansagra & Vaughn, 2013). Untreated, this common disease is associated with a range of cognitive and cardiovascular morbidities (Garetz, 2018).

Various etiological factors underlie pediatric OSA, but the most common is adenotonsillar hypertrophy (Villa et al., 2014). Several medical and surgical treatments exist for OSA, including adenotonsillectomy (A&T), which involves removal of the tonsils and adenoids. An adenotonsillectomy is considered the front line of treatment for otherwise healthy, non-overweight children (in obese children weight loss is often initially prescribed) (Garetz, 2018). Still, as Garetz (2008) discusses, any surgical procedure requires careful risk-benefit and case-by-case analysis to determine if that is the best course of action for the patient as there is risk of complications or persistent disease.

Studies have shown that following an adenotonsillectomy (A&T), most if not all children show improvements in their OSA and sleep-disordered breathing behaviours, but only some find complete

relieve (Villa et al., 2014; Guilleminault et al., 2013; Lee, Guilleminault, Chiu & Sullivan, 2015) Others continue to experience residual OSA symptoms.

Orofacial myofunctional therapy (OMT) is a niche therapy field emerging in North America which involves working with one's craniofacial structures and musculature in order to treat any orofacial dysfunction contributing to breathing, swallowing, speech, feeding, and sleep difficulties. Speech-Language Pathologists can train in this field in order to gain a better understanding of the rehabilitation of the orofacial anatomy. Theoretically, in children with OSA secondary to adenotonsillar hypertrophy, the removal of those bulky tissues via A&T can cause problems with muscle recruitment, as structures will have to re-learn their motor patterns in order to restore proper function now that structures are different and there is more space. OMT may be used to facilitate this muscle re-education.

### ***Objectives***

The primary objective of this paper was to critically review existing literature in order to gain an understanding of the role orofacial myofunctional therapy may have as a treatment tool for children with obstructive sleep apnea.

### ***Methods***

#### Search Strategy

A variety of computerized databases, including PubMed, Scopus, Web of Science, and Google Scholar, were searched with the following search terms: (orofacial myofunctional therapy) OR (OMT) AND (sleep apnea) OR (sleep disordered breathing) AND (children). Search was limited to peer-reviewed journal articles written in English. Reference lists of searched articles also used.

#### Selection Criteria

Articles were included in this review if they directly addressed the efficacy and impact of OMT on the

treatment of OSA. Studies were limited to pediatric subjects (defined as at or under the age of 10) who were not obese, and articles that evaluated the efficacy of external assistive devices (i.e. an oral device worn during sleep) were excluded.

#### Data Collection

The results of the literature search yielded four studies that met inclusion criteria: one randomized control trial, two retrospective chart reviews, and one prospective case control study.

### *Results*

**Guilleminault et al, 2013** conducted a retrospective chart review (level of evidence = 2C) designed to evaluate the impact of OMT on orofacial muscle weakness following surgical (A&T) and orthodontic interventions for sleep disordered breathing. Children aged 3.6 - 6.6 at initial visit that met all inclusion criteria and whose charts had necessary follow-up information were included in the study (n = 24). Following A&T and/or palatal expansion, participants with residual OSA were referred to orthodontic care and to OMT; 11 children received OMT and 13 did not.

Use of OMT by subjects was documented as completed as per recommendations or not followed, and subjects were evaluated at follow-up two years and four years after OMT care was terminated. All 13 subjects that did not receive OMT showed a recurrence of OSA symptoms. In contrast, all 11 subjects that completed OMT for two years post-surgery achieved did not demonstrate a recurrence of symptoms.

Though the rationale for the study was clear and the study design was well-formulated and replicable, participant inclusion criteria were not clearly explained. However, follow-up over a long-term period was extensive, and objective comparative data was collected at each in the form of a nocturnal polysomnography (PSG), which is a strength of this study. Though the initial diagnostic PSGs were not available to review and researchers had to rely only on the report on file, the long-term follow-up PSGs were scored by a single-blinded scorer to avoid interlaboratory and interscorer variability. Results from PSGs from subjects who had and who had not received OMT were compared using Wilcoxon signed-rank test and Chi squared tests.

Though having long-term follow-up data is beneficial, it required charts to have specific initial and follow-up data reported clearly enough to analyze. These restrictions resulted in a small participant group, which is a limitation of this study. Additionally, because data was obtained from three different locations, medical records were not easily retrievable, likely contributing to the small number of participants. These different locations could have

created variability in the data collection and results, and because of this, there was also not a standardized treatment protocol followed by the various OMT specialists seen by participants, though authors report that protocols were similar. Authors also state that only those children that complied with their OMT protocols were included in the OMT group.

Overall, this study provides suggestive evidence that OMT may be a useful treatment that significantly decreases risk of the recurrence of OSA in children post-adenotonsillectomy.

**Villa et al., 2014** evaluated the efficacy of oropharyngeal exercises in children with OSA after A&T by conducting a randomized control trial (level of evidence = 1). Children included in this study were aged 4.4 – 7.6. Clinical history was obtained for all patients, and all underwent an ear, nose and throat (ENT) and an orthodontic assessment. Polysomnographic recordings (PSGs) were performed before A&T and were repeated six months later. At the six-month point, patients with residual OSA following A&T (n = 30 out of the original 42) were randomized into two groups (T1). Group 1 included 14 participants and was considered the experimental group, and Group 2 contained 13 participants and was the control group. Two months later (T2; therefore, 8 months after A&T), participants were re-evaluated using PSG and clinical evaluation. Three participants with residual OSA were excluded due to non-compliance with the exercise protocol (n = 2), or for taking nasal steroids in addition to control group protocol (n = 1). Interestingly, 88.8% of participants with residual OSA were male.

There were no significant differences between groups' demographics, clinical findings, nor in the AHI (Apnea Hypopnea Index; or the number of apnea or hypoxic events per hour) at T1. The authors reveal that after two months of completing oropharyngeal exercises (protocol not specified), participants in the experimental Group 1 has a significant reduction in oral breathing, increased labial seal and lip tone, and demonstrated a significantly larger difference in  $\Delta$ AHI from T1 to T2 (Group 1 = 58.01%, Group 2 = 6.96%).

This study also used the Glatzel Test and the Rosenthal Test to evaluate nasal patency. Upon investigation, it seems that both of these tests are not validated in current literature, nor are they commonly used in practice anymore.

A T-test was conducted for the parametric data, and a Chi squared test was used to compare the data. Improvements in OSA were defined by the difference in the Apnea-Hypopnea Index ( $\Delta$ AHI). The authors concluded that the AHI is significantly reduced in patients that used an

oropharyngeal exercise protocol following orthodontic and/or surgical intervention in the form of an A&T.

Strengths of this study include its randomized design, the clearly described hypothesis, and that the inclusion and exclusion criteria were reported in detail. Data was collected prospectively, and the main findings of the study were related to initial outcomes and were clearly defined.

Limitations of this study include the use of unvalidated non-standardized tests (Rosenthal and Glatzel tests) to evaluate nasal patency, though fortunately more in-depth and objective measures were used to evaluate and interpret results. There is a relatively small sample size, likely due to the frequency of the surgery in this age group, and strict exclusion criteria. Though authors stated there was varying degrees of participant compliance, data regarding exercise compliance is not reported.

Overall, this article presents suggestive evidence that oropharyngeal exercises, which are part of an OMT's purview, are valuable to children post-A&T in reducing the recurrence of OSA symptoms and oral breathing.

**Lee et al., 2015** conducted a retrospective chart review (level of evidence = 2C) to investigate whether myofunctional re-education was effective to alter the mouth breathing pattern in children, and if this can have an impact on symptoms of sleep-disordered breathing. They also investigated the frequency of oral breathing during sleep before and after an A&T. Authors defined mouth breathing during sleep as a minimum of 35% of total sleep time. Mouth breathing is associated with adenotonsillar hypertrophy, which is a primary cause of pediatric OSA (SC et al., 2010).

In the initial cohort evaluated were 64 children between the ages of 3 and 9. All children underwent surgical intervention via A&T and thus experienced significant improvements in OSA symptoms. Before A&T, the mean AHI was  $8.58 \pm 3.15$  events per hour, and after surgical intervention, the PSG revealed that the mean AHI dropped to  $1.71 \pm 1.21$ . In 29 subjects, the AHI after surgery was  $>1.50$ , which the authors defined as residual OSA if there was also the presence of overall symptoms (i.e. fatigue, snoring, inattention, hyperactivity). Of the 64, 35 children still displayed evidence of mouth breathing following A&T (44-100% of total sleep time). Some of the children who were mouth breathers did not display symptoms of OSA ( $n = 9$ ). All subjects with persistent mouth breathing ( $n = 35$ ) were given myofunctional exercises from online sources to perform for 6 months and were referred to OMT specialists.

Of the 35 subjects referred to OMT, 29 returned for follow-up evaluation 6 months later, and repeat PSG one year later. Just 7 of these participants reported receiving

OMT services. Education was provided regarding the importance of the exercises to all returning subjects ( $n = 29$ ) and additional OMT referrals were provided.

At the one-year post-A&T follow-up, 18 children in the mouth breathing group underwent evaluation and another PSG. In this subgroup, 9 children had received OMT services. The authors used Chi squared and T-tests to compare between-group data. The children who received OMT ( $n = 9$ ) versus those who had not ( $n = 9$ ) at the 1-year follow-up showed improvements in AHI, oxygen saturation, and nasal flow limitation. A repeated measures analysis using general linear modelling was used for AHI, flow limitation and oxygen saturation measures.

The authors conclude that the assessment of mouth breathing during sleep should be performed in children with sleep disordered breathing, and also after A&T. It is stated that the persistence of mouth breathing should be treated with OMT. The authors clearly state inclusion and exclusion criteria, and the objectives and reasoning for them are described. The study design was tailored to the objectives, and assessments were set up for long-term follow-up. However, follow-up data is limited, which is a major limitation of this study. At one year, only 9 children of the original cohort had completed OMT. Additionally, this is not a randomized study, and we were not informed as to which exercises were used in the OMT protocol, the similarities and differences between protocols used by participants (as there were three different therapists), the frequency of administration, nor compliance data. Though different surgeons completed the A&T surgeries, which is a limitation, the same rater scored the pre- and post- PSGs, functioning to reduce inter-rater variability.

Overall, this article presents suggestive evidence of the efficacy of OMT in reducing symptoms of OSA in children.

**Villa et al., 2017** investigated the role of OMT in increasing tongue tone, and subsequent effects in children with sleep-disordered breathing. The authors used a prospective case control study (level of evidence = 2A) with 54 total participants randomly assigned to Group 1 ( $n = 36$ ) who underwent an OMT protocol, and Group 2 ( $n = 18$ ) who did not. A control group ( $n = 38$ ) of age- and sex-matched controls (non-obese, no history of sleep or respiratory problems) was randomly recruited.

PSG was used before treatment to determine the severity of the sleep-disordered breathing, and OSA was diagnosed in participants if they had an AHI greater than or equal to 1 event per hour, combined with symptoms of OSA (i.e. fatigue, inattention, hyperactivity, snoring). Participants in both groups displayed mild OSA. Assessments included lip and tongue strength/endurance

measures using the IOPI (an evidence-based validated tool with norms used for measuring tongue and lip strength and endurance (Adams, Mathisen, Baines, Lazarus & Callister, 2013), PSG, myofunctional clinical evaluation and pulse oximetry. Both the OMT and the non-OMT group underwent all assessments initially and after two months, and the control group underwent only IOPI measurements.

The authors define the myofunctional treatment as “isometric and isotonic exercises involving the tongue, soft palate, and lateral pharyngeal walls, designed to improve suction, swallowing, chewing breathing and speech functions” (p.1027). Participants were told to perform the exercises three times daily, with 10-20 repetitions each time. One therapist administered all clinical OMT evaluations and prescribed all treatment plans to patients in an effort to reduce observer bias and inter-therapist variability.

The Wilcoxon signed rank test was used to compare data from before and after OMT, and Chi squared and T-tests were conducted for continuous variables. Before treatment, when compared to the healthy control group, the OMT and non-OMT groups had reduced tongue strength, peak, and longer endurance (seconds), and the authors hypothesize that this is likely due to oral breathing when sleeping, which affects tongue position and strength, ultimately leading to abnormal development of craniofacial and airway structures. Following treatment, the OMT group showed significantly increased tongue strength, peak, and endurance, lip tone, oxygen saturation, and a decrease in their oral breathing habit. There were no differences in the non-OMT group.

The aim and hypothesis were clearly described, and inclusion and exclusion criteria were reported clearly. The method was duplicable, which is among the strengths of this study. The randomization of participants to groups is a strength, as is the relatively large sample size.

Additional limitations of this study include that PSGs were not repeated following treatment to avoid high costs and because parents were satisfied with improvements and did not require a repeat examination. Only IOPI measurements were completed. The study only included participants with mild-to-moderate OSA, and the adherence to OMT treatment protocol was not defined. The study follows participants for just two months, which clearly was enough to demonstrate change, but not enough time for authors to make any conclusions about OMT's long term effects. Finally, it does not appear that the authors reported any comparison between the OMT group and the control group following treatment.

Overall, this study provides suggestive evidence that OMT can be beneficial in decreasing symptoms of and improving outcomes for children with OSA.

### *Discussion*

Pediatric obstructive sleep apnea occurs when a child stops breathing for periods during sleep. The common cutoff for OSA diagnosis in children is an AHI  $\geq 1$ , when also combined with symptoms of OSA. In the review of this literature, three articles used this cutoff (Guilleminault et al., 2013; Villa et al., 2014; Villa et al., 2017), whereas Lee et al. (2015) defined residual OSA post A&T as AHI  $\geq 1.5$ , which means that some participants that were included in their non-residual group may have had persistent symptoms.

Orofacial myofunctional therapists work to assess and treat disordered orofacial and pharyngeal musculature in order to improve patient's suck, swallow and breathing habits as well as at rest postures. It is a relatively small niche practice, and currently dentists, dental hygienists and Speech-Language Pathologists are equipped to train in this field. The literature described in this paper all had high levels of evidence and cumulatively suggest that OMT can play a role in the treatment of obstructive sleep apnea in children.

The literature discussed (with the exception of Villa et al. (2017)) involved patients who had previously undergone an adenotonsillectomy. The primary clinical indication for A&T in children with OSA is when the tonsils and adenoids are relatively large compared to the size of the child's airway and are occlusive.

Additionally, as discussed, obesity may be a cause of sleep apnea (CL et al., 2012), which is why they were excluded from experimental and/or control groups (Guilleminault et al., 2013; Villa et al., 2014; Lee et al., 2015; Villa et al., 2017). In all, just Villa et al. (2017) had a participant pool that had not undergone surgical intervention. A&T is still considered the gold standard treatment in children with OSA

A general trend in the literature was the lack of reporting of compliance to the OMT protocol. Only one paper discussed frequency of administration (Villa et al., 2017), and Villa et al., (2014) indicated that they excluded participants' data due to non-compliance.

Another general trend was that the OMT protocol itself was not reported. In general, the authors presented what the protocol's focus was, but never specific exercises within a regimen. In all but one (Villa et al., 2017), there were different orofacial myofunctional therapists administering treatment protocols, which allowed for them to be blinded, but contributed to high variability.

A weakness of the total literature pool is the lack of consistent terminology. Many terms were used to discuss OMT: orofacial myofunctional therapy, myofascial re-education (Guilleminault et al., 2013), myofunctional re-education (Lee et al., 2015), myology, myofunctional therapy (Villa et al., 2017), oropharyngeal exercises. This inconsistency will lend itself only to confuse readers and researchers. Compiling literature based on consistent terminology will be important to this type of practice. Additionally, this inconsistency can be cited as a weakness of this literature review, as search terms may not have captured all relevant articles.

Taken together, the results of the four reviewed articles provide promising evidence that orofacial myofunctional therapy can be beneficial for children experiencing obstructive sleep apnea. There is more research to demonstrate its effectiveness in minimizing residual OSA following surgical intervention, and little regarding the effectiveness of OMT on mild OSA without surgery.

### **Conclusion**

It is clear that orofacial myofunctional therapy is a niche therapy that has limited research completed. Available literature is current and generally has a high level of evidence but contain marked weaknesses and trend toward under-reporting compliance and OMT protocols. As de Felicia et al. (2018) states, randomized high-quality studies are rare, and OMT needs to be evaluated on a long-term basis. In addition to more long-term follow-up studies, more research needs to be completed on different subject groups; i.e. pre-pubertal versus pubertal participants, those with normal weight versus obese, and those with varying severities of OSA with or without surgery.

### **Clinical Implications**

Orofacial myofunctional therapy is an up-and-coming practice in North America, and based on current research, it seems to be based in evidence for treating children with OSA following surgical intervention, for muscular re-education. When certified via the Academy of Orofacial Myofunctional Therapy (AOMT) or the International Association of Orofacial Myology (IAOM) and practicing, clinicians must be aware of new research published and be guarded about certain things that are not well understood. For example, it is not well understood what dose of therapy should be recommended for maximal benefits in children who have had/not had surgical intervention, or who have varying degrees of severity of OSA. Additionally, certain groups of patients are likely able to tolerate this type of exercise therapy more effectively, and Guilleminault et al. (2013) state

that re-education is likely easier in children ages six and older because of the degree of effort, understanding, and compliance it takes to perform the exercises.

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