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Myofunctional Therapy to Treat Obstructive Sleep Apnea: A Systematic Review and Meta-analysis

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Objective: To systematically review the literature for articles evaluating myofunctional therapy (MT) as treatment for obstructive sleep apnea (OSA) in children and adults and to perform a meta-analysis on the polysomnographic, snoring, and sleepiness data.

Data Sources: Web of Science, Scopus, MEDLINE, and The Cochrane Library.

Review Methods: The searches were performed through June 18, 2014. The Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) statement was followed.

Results: Nine adult studies (120 patients) reported polysomnography, snoring, and/or sleepiness outcomes. The pre- and post-MT apnea-hypopnea indices (AHI) decreased from a mean ± standard deviation (M ± SD) of 24.5 ± 14.3/h to 12.3 ± 11.8/h, mean difference (MD) −14.26 [95% confidence interval (CI) −20.98, −7.54], P < 0.0001. Lowest oxygen saturations improved from 83.9 ± 6.0% to 86.6 ± 7.3%, MD 4.19 (95% CI 1.85, 6.54), P = 0.0005. Polysomnography snoring decreased from 14.05 ± 4.89% to 3.87 ± 4.12% of total sleep time, P < 0.001, and snoring decreased in all three studies reporting subjective outcomes. Epworth Sleepiness Scale decreased from 14.8 ± 3.5 to 8.2 ± 4.1. Two pediatric studies (25 patients) reported outcomes. In the first study of 14 children, the AHI decreased from 4.87 ± 3.0/h to 1.84 ± 3.2/h, P = 0.004. The second study evaluated children who were cured of OSA after adenotonsillectomy and palatal expansion, and found that 11 patients who continued MT remained cured (AHI 0.5 ± 0.4/h), whereas 13 controls had recurrent OSA (AHI 5.3 ± 1.5/h) after 4 y.

Conclusion: Current literature demonstrates that myofunctional therapy decreases apnea-hypopnea index by approximately 50% in adults and 62% in children. Lowest oxygen saturations, snoring, and sleepiness outcomes improve in adults. Myofunctional therapy could serve as an adjunct to other obstructive sleep apnea treatments.

Keywords: exercise therapy/methods, myofunctional therapy/methods, obstructive sleep apnea, sleep apnea syndromes

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INTRODUCTION

Several medical and surgical treatment modalities exist as treatment for obstructive sleep apnea (OSA).[1–3] Four pathophysiological traits seen in patients with OSA are: the passive critical closing pressure of the upper airway (Pcrit), arousal threshold, loop gain, and muscle responsiveness (PALM) with categories of 1, 2, 2a, 2b, and 3.[4] It has been demonstrated that patients in four of five PALM categories will benefit from anatomic interventions.[4] Because the dilator muscles of the upper airway play a critical role in maintaining an open airway during sleep, researchers have explored exercises and other airway training (singing, didgeridoo, instrument playing) that target oral cavity and oropharyngeal structures as a method to treat OSA.[5–7] Myofunctional therapy (MT) and proper tongue positioning in the oral cavity have been described since 1918 to improve mandibular growth, nasal breathing, and facial appearance.[8] Guimaraes has proposed MT as a treatment for OSA since the 1990s.[9] MT is composed of isotonic and isometric exercises that target oral (lip, tongue) and oropharyngeal structures (soft palate, lateral pharyngeal wall).[7,10] There have been an increasing number of studies evaluating the effect of MT in the form of case studies, case series, and most recently, two randomized controlled trials.[7,10–13]

The most comprehensive MT exercises are described by Guimaraes et al.[7] and involve the soft palate, tongue, and facial muscles and address stomatognathic functions. For soft palate exercises, patients pronounce oral vowel sounds either continuously (isometric exercises) or intermittently (isotonic exercises).[7] Tongue exercises include moving the tongue along the superior and lateral surfaces of the teeth, positioning the tongue tip against the anterior aspect of the hard palate, pressing the entire tongue against the hard and soft palate, and forcing the tongue onto the floor of the mouth.[7] Facial exercises address the lip (i.e., contraction and relaxation of the orbicularis oris), buccinators (i.e., suction movements and application of intraoral finger pressure against the buccinator muscles), and jaw muscles (i.e., lateral jaw movements).[7] In addition, stomatognathic functions are addressed by instructing patients to inhale nasally and exhale orally without and then with balloon inflation, and performing specific swallowing and chewing exercises (i.e., swallowing with the teeth clenched together, tongue positioned in the palate and without contraction of

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perioral muscles; alternating chewing sides).[7] A newer study describes a device that conditions and strengthens oral and tongue muscles.[12]

The objective of this study is to systematically review the literature for articles evaluating MT or oral/oropharyngeal exercises as treatment for OSA in both children and adults and to perform a meta-analysis on the available polysomnographic and sleepiness data.

METHODS

Search Strategy

A search was performed on Web of Science, Scopus, MEDLINE, and The Cochrane Library, initially January 18, 2014, with an update on June 18, 2014. MeSH terms and keywords used for the search included various combinations of the following: "myofascial reeducation," "myofunctional therapy," "obstructive sleep apnea," "orofacial myotherapy," "oral myotherapy," "oropharyngeal exercises," "sleep," "sleep apnea syndromes," "speech therapy," "upper airway exercises," and "upper airway remodeling." One example of a MEDLINE search is: ((("Myofunctional Therapy"[MeSH]) AND "Sleep Apnea Syndromes"[MeSH])) OR ("sleep" AND ("myofascial reeducation" OR "myofunctional therapy" OR "orofacial myotherapy" OR "oral myotherapy" OR "oropharyngeal exercises" OR "speech therapy" OR "upper airway exercises" OR "upper airway remodeling")).

For each of the searches, the titles and abstracts were screened and the full text versions of articles that met criteria were downloaded. Full texts were reviewed and any referenced articles that were not already obtained were ordered and obtained. "Related citations" were also reviewed during the searches, and the "cited by" function on Google Scholar was also used to identify any additional studies.

Study Selection

Criteria for inclusion included peer-reviewed studies (published articles or abstracts) evaluating oral or oropharyngeal MT as an isolated treatment for either adult or pediatric OSA; studies needed to report quantitative polysomnographic, snoring, and/or sleepiness data pretreatment and posttreatment or they needed to report the percentage of difference between pretreatment and posttreatment outcomes. All languages were included. Exclusion criteria included studies evaluating singing, instrument playing, and studies without quantitative data. If individual patient data were reported and patients lost 10% or more of their body weight, then those patients were excluded. Studies in which the MT patients also underwent additional interventions such as continuous positive airway pressure therapy, mandibular advancement device therapy, sleep apnea surgery, allergy management, weight loss management, or any other intervention that could also contribute to improved sleep apnea outcomes were excluded (unless the additional interventions were performed in control groups and the data were provided separately for both MT and control groups).

Data Abstraction and Study Quality Assessment

Authors MC, JA, and SZ independently performed a search of the literature and screened titles and abstracts and downloaded the articles for inclusion. The decision to include the articles was made by consensus, and if necessary the final decision was made by author MC. Data collected included patient age, body mass index (BMI), polysomnographic data (AHI, lowest oxygen saturation), snoring, and sleepiness data. If data were missing from the articles, then the corresponding author was contacted in an attempt to obtain the data. The corresponding author of the study by Suzuki et al.[12] was contacted and confirmed that the reported oxygen saturation data were for lowest oxygen saturation and that tongue training was involved as part of the MT device training.

The National Institute for Health and Clinical Excellence (NICE) quality assessment tool was used to evaluate the quality of the included studies. The instrument consists of eight items that are assessed for each individual study.

Statistical Analysis

The statistics were performed with the IBM Statistical Package for Social Sciences software (SPSS) version 20.0 (Armonk, New York, USA). Means and standard deviations were calculated before and after myofunctional therapy. Studies providing raw patient data without means and standard deviations were manually input into SPSS for calculation; or if individual scatterplots with pretreatment and posttreatment data were available, the estimated data point values were used to calculate the means and standard deviations. The null hypothesis for this study is that there is no difference in outcome data before and after myofunctional therapy. For combining data, a two-tailed, paired t test was performed ($P < 0.05$ was the cutoff for significance). Review Manager (RevMan) [Computer program] Version 5.3 (Copenhagen: The Nordic Cochrane Centre, The Cochrane Collaboration, 2014) was used for meta-analysis. A random-effects model was used if heterogeneity existed and a fixed-effects model was used if no heterogeneity existed. When pooling the data in studies, the means, standard deviations, and 95% confidence intervals (CI) were calculated by REVMAN. Heterogeneity was assessed by I^2 statistic (inconsistency levels: low = 25%, moderate = 50% and high = 75%)[14] and the Cochran Q statistic (with significant heterogeneity being considered when $P \leq 0.1$ was obtained).[15] If heterogeneity existed, then a sensitivity analysis was performed by removing each of the studies individually to identify the source(s).

The Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines were downloaded and followed during this review.[16]

RESULTS

A total of 226 studies were screened for relevance, and 204 were excluded. After identification of 22 potentially relevant studies, they were downloaded and the reviews of the reference lists yielded an additional 6 studies, for a total of 28 studies.[7–13,17–36] Nine were review articles,[8,20,22,27,30,32–34,36] two reported no intervention,[24,31] two studied lip exercises and the effect on lip thickness,[21,37] one reported breathing exercises not involving oral cavity or oropharyngeal structures,[28] one was a letter to the editor,[11] and two studies were abstracts in which data were later reported in the authors' journal articles.[19,25] Eleven studies met criteria and were included in this review. Individual patient data were reported by one pediatric study[35] and one adult study,[12] whereas the remaining nine studies reported outcomes with means and standard deviations.[7,9,10,13,17,18,23,26,29] Figure 1 summarizes the flow for study selection.

Methodological Quality of the Included Studies

The studies included in this review included one abstract,[26] one retrospective case report,[23] three retrospective case series,[9,10,18] three prospective case series,[12,17,29] one randomized trial,[35] and two randomized controlled trials.[7,13] Most of the studies satisfied four to six of the eight NICE quality assessment tool items (presented in Table S1 of supplemental material). The main limitations were that the total number of patients in most studies was low, the studies were at single institutions (except one that was multicentered) and most studies did not explicitly state that patients were consecutive.

Adult Studies

A total of nine adult studies (120 patients, age 44.5 ± 11.6 y, BMI 28.9 ± 6.2 kg/m^2) reported polysomnography and/or sleepiness outcomes (Table 1). Baz et al.[17] reported using American Academy of Sleep Medicine (AASM) scoring criteria but did not specify which year, Diaferia et al.[13] and Guimaraes et al.[7] reported using 1999 AASM scoring criteria, Suzuki et al.[12] scored based on the 2005 update to AASM scoring criteria, and the remaining five studies did not specify which polysomnography scoring criteria were used.[9,18,23,26,29]

The pre- and post-MT AHI mean ± standard deviation (M ± SD; 82 patients) decreased from 24.5 ± 14.3/h to 12.3 ± 11.8/h, with a mean difference (MD) of −14.26 [95% CI −20.98, −7.54], Z score of 4.16 ($P < 0.0001$) (Figure 2). Both the I^2 statistic (91%) and the Q statistic (value of < 0.00001) demonstrated significant heterogeneity; therefore, studies were individually excluded to identify the source(s). Exclusion of the studies by Suzuki et al.[12] and Berreto et al.[18] resulted in no heterogeneity in the remaining 73 patients, with the I^2 statistic = 0% and the Q statistic value of 0.6. The mean difference for the remaining studies was −10.49 26 [95% CI −12.67, −8.31]. In adult studies in which MT was performed for at least 3 mo, the mean AHI reduced from 25.2 ± 14.6/h to 12.6 ± 12.2/h, which is a 50% reduction.

The lowest oxygen saturation improved in 82 patients from 83.9 ± 6.0% to 86.6 ± 7.3%, MD of 4.19 [95% CI 1.85, 6.54], with an overall Z score of 3.5 ($P = 0.0005$); see Figure 3. Both the I^2 statistic (59%) and the Q statistic (value of 0.05)

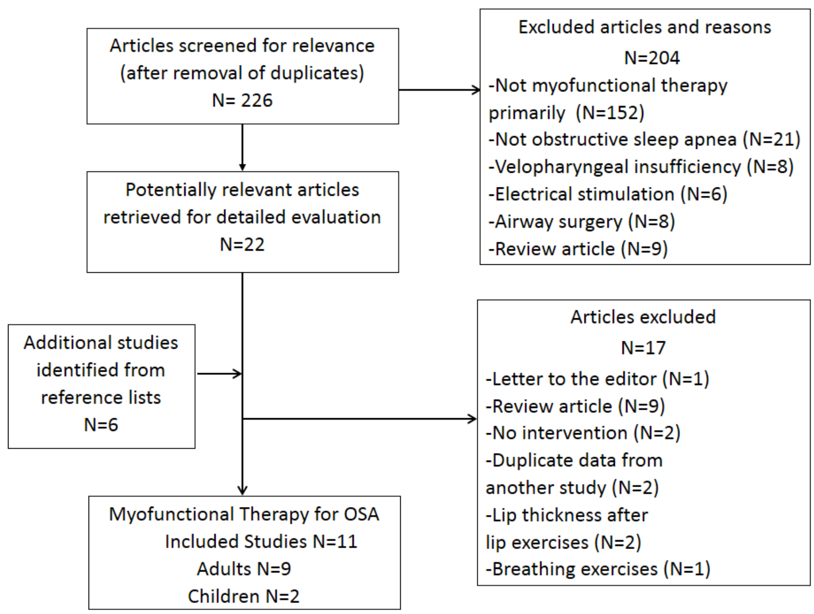

Figure 1—Flow diagram demonstrating myofunctional therapy for obstructive sleep apnea (OSA) study selection. N, number of articles.

Table 1—Adult pre- and post-myofunctional therapy outcomes.

Authors, Year	Study Design	N	Age (years)	BMI (kg/m²)	AHI (events/h) Pre-MT	AHI (events/h) Post-MT	low O_2 (%) Pre-MT	low O_2 (%) Post-MT	ESS Pre-MT	ESS Post-MT
Suzuki et al., 2013*	PCS	6	22.0 ± 0.5	23.8 ± 1.8	15.1 ± 3.4	9.2 ± 1.5	90.0 ± 2.9	96.8 ± 0.8	–	–
Kronbauer et al., 2013	PCS	8	(40–65)	–	–	–	–	–	11.75	4.25
Diaferia et al., 2013	RCT	27	45.2 ± 13.0	25.0 ± 7.4	28.0 ± 22.7	13.9 ± 18.5	83.7 ± 7.7	84.9 ± 8.8	13.7 ± 3.2	7.5 ± 3.7
Baz et al., 2012	PCS	30	44.1 ± 7.5	33.6 ± 2.0	22.3 ± 4.5	11.5 ± 5.4	84 ± 4	87 ± 5	16.4 ± 2.0	9.3 ± 2.9
Guimaraes et al., 2009	RCT	16	51.5 ± 6.8	29.6 ± 3.8	22.4 ± 4.8	13.7 ± 8.5	83 ± 6	85 ± 7	14 ± 5	8 ± 6
de Paula Silva et al., 2007	RCR	1	60	23.3	44	3	83	92	–	–
Berreto et al., 2007	RCS	2	46 ± 12.7	24.2 ± 2.9	44.5 ± 5.7	6.0 ± 3.7	78 ± 1.4	85 ± 2.8	12.5 ± 0.7	4.5 ± 3.5
Guimaraes et al., 2003	ABS	10	–	–	36.1	11.3	–	–	11	7.6
Guimaraes et al., 1999	RCS	20	(33–55)	–	–	−48%	–	–	–	–
Total		120	44.5 ± 11.6	28.9 ± 6.2	24.5 ± 14.3	12.3 ± 11.8	83.9 ± 6.0	86.6 ± 7.3	14.8 ± 3.5	8.2 ± 4.1

*Study authors confirmed the reported oxygen saturation data was for lowest oxygen saturation. –, not reported, %, percent; ABS, abstract; AHI, apnea-hypopnea index; BMI, body mass index; ESS, Epworth Sleepiness Scale; events/h, events per hour; kg/m^2, kilograms per meter squared; low O_2, lowest oxygen saturation; MT, myofunctional therapy; N, number of myofunctional therapy patients in the study; PCS, prospective case series; RCR, retrospective case report; RCS, retrospective case series; RCT, randomized controlled trial.

Study or Subgroup	Post-Myofunctional Tx Mean	SD	Total	Pre-Myofunctional Tx Mean	SD	Total	Weight	Mean Difference IV, Random, 95% CI	Year
Suzuki 2013	9.2	1.5	6	15.1	3.4	6	23.3%	-5.90 [-8.87, -2.93]	2013
Diaferia 2013	13.9	18.5	27	28	22.7	27	14.7%	-14.10 [-25.15, -3.05]	2013
Baz 2012	11.5	5.4	30	22.3	4.5	30	23.7%	-10.80 [-13.32, -8.28]	2012
Guimaraes 2009	13.7	8.5	16	22.4	4.8	16	21.8%	-8.70 [-13.48, -3.92]	2009
Berreto 2007	6	3.7	2	44.5	5.7	2	16.5%	-38.50 [-47.92, -29.08]	2007
Total (95% CI)			**81**			**81**	**100.0%**	**-14.26 [-20.98, -7.54]**	

Heterogeneity: Tau² = 48.01; Chi² = 43.66, df = 4 (P < 0.00001); I² = 91%
Test for overall effect: Z = 4.16 (P < 0.0001)

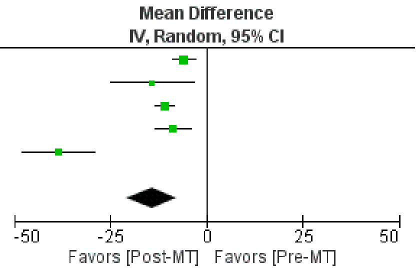

Figure 2—Adult premyofunctional and postmyofunctional therapy outcomes for apnea-hypopnea index (events per hour). CI, confidence interval; MT, myofunctional therapy; SD, standard deviation; Tx, treatment.

Study or Subgroup	Post-Myofunctional Tx Mean	SD	Total	Pre-Myofunctional Tx Mean	SD	Total	Weight	Mean Difference IV, Random, 95% CI	Year
Suzuki 2013	96.8	0.8	6	90	2.9	6	26.0%	6.80 [4.39, 9.21]	2013
Diaferia 2013	84.9	8.8	27	83.7	7.7	27	15.8%	1.20 [-3.21, 5.61]	2013
Baz 2012	87	5	30	84	4	30	26.7%	3.00 [0.71, 5.29]	2012
Guimaraes 2009	85	7	16	83	6	16	15.4%	2.00 [-2.52, 6.52]	2009
Berreto 2007	85	2.8	2	78	1.4	2	16.1%	7.00 [2.66, 11.34]	2007
Total (95% CI)			**81**			**81**	**100.0%**	**4.19 [1.85, 6.54]**	

Heterogeneity: Tau² = 4.00; Chi² = 9.74, df = 4 (P = 0.05); I² = 59%
Test for overall effect: Z = 3.50 (P = 0.0005)

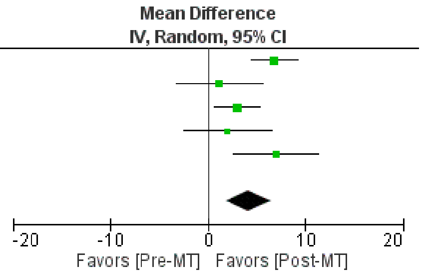

Figure 3—Adult premyofunctional and postmyofunctional therapy outcomes for lowest oxygen saturation (percent). CI, confidence interval; MT, myofunctional therapy; SD, standard deviation; Tx, treatment.

Study or Subgroup	Post-Myofunctional Tx Mean	SD	Total	Pre-Myofunctional Tx Mean	SD	Total	Weight	Mean Difference IV, Fixed, 95% CI	Year
Diaferia 2013	7.5	3.7	27	13.7	3.2	27	28.5%	-6.20 [-8.05, -4.35]	2013
Baz 2012	9.3	2.9	30	16.4	2	30	61.0%	-7.10 [-8.36, -5.84]	2012
Guimaraes 2009	8	6	16	14	5	16	6.6%	-6.00 [-9.83, -2.17]	2009
Berreto 2007	4.5	3.5	2	12.5	0.7	2	4.0%	-8.00 [-12.95, -3.05]	2007
Total (95% CI)			**75**			**75**	**100.0%**	**-6.81 [-7.79, -5.82]**	

Heterogeneity: Chi² = 1.02, df = 3 (P = 0.80); I² = 0%
Test for overall effect: Z = 13.55 (P < 0.00001)

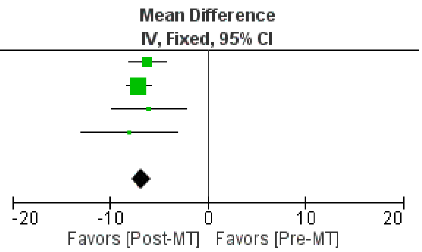

Figure 4—Adult premyofunctional and postmyofunctional therapy outcomes for Epworth Sleepiness Scale. CI, confidence interval; MT, myofunctional therapy; SD, standard deviation; Tx, treatment.

demonstrated significant heterogeneity, therefore, studies were individually excluded to identify the source(s). Exclusion of the studies by Suzuki et al.[12] and Berreto et al.[18] resulted in no heterogeneity in the remaining 73 patients, with the I^2 statistic = 0% and the Q statistic value of 0.56. Oxygen desaturation index was reported by one study, and demonstrated a reduction from 14.53 ± 5.04 to 9.27 ± 4.27, pre- and post-MT, respectively.[17] Sleepiness decreased in all studies reporting the outcome. The Epworth Sleepiness Scale (ESS)[38] decreased in 75 patients from 14.8 ± 3.5 to 8.2 ± 4.1, MD of −6.81 [95% CI −7.79, −5.82], with an overall Z score of 13.55 ($P < 0.00001$); see Figure 4. Both the I^2 statistic (0%) and the Q statistic (value of 0.8) demonstrated no heterogeneity.

Snoring

Snoring changes were evaluated by 4 studies, Baz et al.,[17] Berreto et al.,[18] de Paula Silva et al.[23] and Guimaraes et al.[7]; see Table 2. Baz et al.[17] reported that 30 patients snored before therapy and 16 snored after therapy, P = 0.008 (yes versus no; article did not specify if patient or bed partner was asked) and the polysomnography demonstrated that the percent of total sleep time spent snoring decreased from 14.05 ± 4.89% to 3.87 ± 4.12% (before and after, respectively), $P < 0.001$.[17] Guimaraes et al.[7] found snoring frequency decreased by 25% (article did not specify if patient or bed partner was asked) from 4 to 3 (based on 0 = never to 4 = everyday), P = 0.001, and the snoring intensity decreased by 66% from 3 to 1 (based on 1 = similar to breathing and 3 = very loud) with P = 0.001; whereas the control groups had no change in snoring frequency or intensity. The case study by de Paula Silva et al.[23] demonstrated a decrease in snoring intensity after 8 sessions. Berreto et al.[18] described two patients who decreased from a (bed partner) snoring score of 3 down to 2 (0 = snoring absence, 1 = heavy breathing, 2 = light snoring, 3 = snoring that disturbs the bed partner and 4 = snoring that can be heard outside the bedroom).

Table 2—Snoring outcomes based on mean values pre and post-myofunctional therapy.

Authors, Year	N	Subjective Snoring		PSG %TST Snoring	
		Pre-MT	Post-MT	Pre-MT	Post-MT
Baz et al., 2012	30	Yes = 30; No = 0	Yes = 16; No = 14	14.05 ± 4.89%	3.87 ± 4.12%
Guimaraes et al., 2009	16	Very loud	Similar to breathing	–	–
de Paula Silva et al., 2007	1	Snoring	Decreased snoring	–	–
Berreto et al., 2007	2	Disturbs bedpartner	Light snoring	–	–

MT, myofunctional therapy; %TST, percentage of total sleep time. Snoring outcomes are based on quantified definitions pre- and post-myofunctional therapy by all studies except de Paula Silva et al. (case report).

Pediatric Studies

A total of two pediatric studies (25 patients, age 8.4 ± 3.1 y) reported polysomnography and/or sleepiness outcomes. Both pediatric studies reported using 2007 AASM scoring criteria, and Guilleminault et al.[10] also specified that hypopneas were scored with a 50% reduction in nasal cannula curve and an associated 3% or more reduction in oxygen saturation and/or with associated arousals, while Villa et al.[35] did not specify the hypopnea scoring criteria. The study by Villa et al.[35] was a prospective randomized controlled trial in which postadenotonsillectomy patients were randomized to either oropharyngeal exercises or control group. The treatment group in this study consisted of 14 patients and the pre- and post-MT AHI was evaluated after 2 mo of oropharyngeal exercises. The AHI M ± SD reduced from 4.87 ± 3.0/h to 1.84 ± 3.2/h, P = 0.004 (a 62% reduction).[35] The control group had minimal change in AHI during the 2-mo period (4.56/h down to 4.11/h).[35] The study by Guilleminault et al.[10] was a retrospective chart review, evaluating 24 children who were cured by the combination of adenotonsillectomy and palatal expansion (AHI 0.4 ± 0.3); and 11 of the children received MT (intervention group) and 13 children did not receive MT (controls).[10] At the 4-y follow-up, the children who practiced MT over the long term remained cured of OSA (AHI 0.5 ± 0.4), compared to children who were never trained to perform the exercises and subsequently had a recurrence of OSA (AHI 5.3 ± 1.5/h).[10] Although both pediatric MT studies compared the intervention groups to control groups, neither study reported pretreatment and posttreatment lowest oxygen saturation or sleepiness outcomes.

DISCUSSION

This systematic review and meta-analysis of nine adult and two pediatric studies evaluating the effect of MT on OSA has five main findings. First, MT provides a reduction in AHI of approximately 50% in adults and 62% in children. The pre- and post-MT AHI for adults decreased from 24.5 ± 14.3/h to 12.3 ± 11.8/h, MD of −14.26 [95% CI −20.98, −7.54] (P < 0.0001). For pediatric patients, the pre- and post-MT M ± SD for AHI decreased from 4.87 ± 3.0/h to 1.84 ± 3.2/h, P = 0.004. Additionally, the study by Guilleminault et al.[10] reported that 11 children remained cured of OSA (AHI of 0.5 ± 0.4/h) after continuing MT for 4 y. There was heterogeneity, and the studies by Suzuki et al.[12] and Berreto et al.[18] were shown to be the sources. The study by Suzuki et al.[12] had six patients, who used an oral exercise device to help train, but the length of time between polysomnography was 2 mo, whereas the remaining adult studies reporting AHI had a follow-up duration of at least 3 mo between polysomnography studies. Had the study been extended to 3 mo, there may have been additional improvement in AHI. In studies with control groups, there was little to no improvement in the AHI for the control groups compared to improvement in the MT intervention group. There is also a clear improvement in lowest oxygen saturation by approximately 3–4%, with the meta-analysis of 81 patients demonstrating a mean difference pre- and post-MT of 4.19%, [95% CI 1.85, 6.54]. The oxygen desaturation index (ODI) was only reported by Baz et al.,[17] demonstrating a 36% reduction, but the article did not specify whether the ODI in the study was based on 3% or 4% desaturation.

Second, MT decreases snoring both subjectively and objectively. Four studies compared the pre- and post-MT outcomes and it was noted that snoring decreased after therapy (three of four studies quantified the snoring). The polysomnography demonstrated a 72.4% reduction in snoring pre- versus post-MT (14.05 ± 4.89% to 3.87 ± 4.12%, before and after, respectively), P < 0.001.[17] With regard to subjective improvement in snoring intensity, the three studies quantifying the outcomes reported that during posttreatment there was a decrease in snoring to either light snoring, or the sound was similar to normal breathing.

Third, subjective sleepiness also improves post-MT as demonstrated by a clear reduction in ESS score for the 93 patients in which it was administered, with a reduction from 14.8 ± 3.5 to 8.2 ± 4.1 (in 75 patients in whom M ± SDs were reported).[7,13,17,18,26,29] The posttreatment ESS is below the threshold for hypersomnia, which is generally considered to be 11 or higher on the scale.[39] Additionally, the 1999 study by Guimaraes[9] reported a subjective reduction in sleepiness; however, the use of a validated sleepiness scale was not specified.[9]

Fourth, despite the heterogeneity in oral and oropharyngeal exercises, overall the improvements in polysomnographic outcomes and sleepiness were consistent. MT was performed for as little as 5 min, twice daily, 4 days a week for 2 mo[12] to as many as 10 min, three to five times daily for 3 mo.[17] The longest published experience with MT for adult OSA has been that of Guimaraes,[9] which is 6 mo. Guimaraes[9] has also published thorough instructions for performing the exercises that involve the soft palate, tongue, facial muscles, and stomatognathic functions to be performed 30 min a day.[7]

Fifth, future research is needed to help explain the pathophysiology and mechanism of action of MT as treatment for

OSA. It can be hypothesized that the exercises improve oral and/or oropharyngeal muscle tone and also may decrease the amount of fatty deposition of the tongue, but this has not been proven. It can be recommended that future researchers consider using the standardized exercises, which have been developed and used over a period of several years by Guimaraes et al.[7] because they have the most experience with the therapy. As pointed out by Guimaraes et al.,[7] because MT is based on an integrative approach with several exercises, it is not possible to determine the effects of specific exercises to determine which ones contribute the most to improvement in outcomes[7]; therefore, future studies could consider exploring the effect of individual exercises. Individual patient data were not available for most studies; therefore, a subanalysis could not be performed for BMI, AHI, age, etc. based on the current literature. However, with regard to BMI, Guimaraes et al.[7] and Baz et al.[17] had significant reductions in AHI in overweight (BMI M ± SD 29.6 ± 3.8) and obese patients (BMI M ± SD 33.6 ± 2.0). With regard to age, the MT has been shown effective in children and adults of all ages studied thus far, ranging from 3 to 60 y.

Limitations

A total of 145 patients (including 25 children) were included in this meta-analysis; however, the magnitude of the effects was highly significant. Although there were nine adult studies, a significant limitation for pediatric studies is that currently only two articles have been published. Additionally, long-term follow-up for more than 6 mo is limited. Except the study by Guilleminault et al,[10] which followed patients for 4 y, all of the other studies spanned 2 to 6 mo. The study by Guilleminault et al.[10] demonstrates a long-term (4 y) maintenance of reduction in AHI and alleviation of OSA symptoms in patients who continued to perform MT exercises, compared to the control group that had recurrence of symptoms and recurrence of an elevated AHI at 4-y follow-up.[10] Because this is the only study that has reported outcomes longer than 6 mo after initiation of MT exercises, additional long-term studies are needed to demonstrate the lasting effects of continued MT. Questions that have not been addressed that could be studied in the future include whether there is a relationship with the tongue exercises and changes in the tongue and palatal muscle tone and/or strength, tongue size (tongue fat), and overall upper airway volume changes pretreatment and posttreatment.

CONCLUSION

Current literature demonstrates that myofunctional therapy decreases AHI by approximately 50% in adults and 62% in children. Lowest oxygen saturation, snoring, and sleepiness outcomes improve in adults. Myofunctional therapy could serve as an adjunct to other OSA treatments.

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REFERENCES

1. Sullivan CE, Issa FG, Berthon-Jones M, Eves L. Reversal of obstructive sleep apnoea by continuous positive airway pressure applied through the nares. Lancet 1981;1:862–5.
2. Camacho M, Certal V, Capasso R. Comprehensive review of surgeries for obstructive sleep apnea syndrome. Braz J Otorhinolaryngol 2013;79:780–8.
3. Randerath WJ, Verbraecken J, Andreas S, et al. Non-CPAP therapies in obstructive sleep apnoea. Eur Respir J 2011;37:1000–28.
4. Eckert DJ, White DP, Jordan AS, Malhotra A, Wellman A. Defining phenotypic causes of obstructive sleep apnea. Identification of novel therapeutic targets. Am J Crit Care Med 2013;188:996–1004.
5. Puhan MA, Suarez A, Lo Cascio C, Zahn A, Heitz M, Braendli O. Didgeridoo playing as alternative treatment for obstructive sleep apnoea syndrome: randomised controlled trial. BMJ 2006;332:266–70.
6. Wardrop PJ, Ravichandran S, Hair M, Robertson SM, Sword D. Do wind and brass players snore less? A cross-sectional study of snoring and daytime fatigue in professional orchestral musicians. Clin Orolaryngol 2011;36:134–8.
7. Guimaraes KC, Drager LF, Genta PR, Marcondes BF, Lorenzi-Filho G. Effects of oropharyngeal exercises on patients with moderate obstructive sleep apnea syndrome. Am J Respir Crit Care Med 2009;179:962–6.
8. Rogers AP. Exercises for the development of muscles of face with view to increasing their functional activity. Dental Cosmos LX 1918;59:857–76.
9. Guimaraes KC. [Soft tissue changes of the oropharynx in patients with obstructive sleep apnea]. J Bras Fonoaudiol 1999;1:69–75.
10. Guilleminault C, Huang YS, Monteyrol PJ, Sato R, Quo S, Lin CH. Critical role of myofascial reeducation in pediatric sleep-disordered breathing. Sleep Med 2013;14:518–25.
11. Steele CM. On the plausibility of upper airway remodeling as an outcome of orofacial exercise. Am J Respir Crit Care Med 2009;179:858–9.
12. Suzuki H, Watanabe A, Akihiro Y, et al. Pilot study to assess the potential of oral myofunctional therapy for improving respiration during sleep. J Prosthodont Res 2013;57:195–9.
13. Diaferia G, Badke L, Santos-Silva R, Bommarito S, Tufik S, Bittencourt L. Effect of speech therapy as adjunct treatment to continuous positive airway pressure on the quality of life of patients with obstructive sleep apnea. Sleep Med 2013;14:628–35.
14. Higgins JP, Thompson SG, Deeks JJ, Altman DG. Measuring inconsistency in meta-analyses. BMJ 2003;327:557–60.
15. Lau J, Ioannidis JP, Schmid CH. Quantitative synthesis in systematic reviews. Ann Intern Med 1997;127:820–6.
16. Moher D, Liberati A, Tetzlaff J, Altman DG. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. PLoS Med 2009;6:e1000097.
17. Baz H, Elshafey M, Elmorsy S, Abu-Samra M. The role of oral myofunctional therapy in managing patients with mild to moderate obstructive sleep apnea. PAN Arab J Rhinol 2012;2:17–22.
18. Berreto e Silva Pitta D, Farias Pessoa A, Sampaio AL, Nonato Rodrigues R, Guiot Tavares M, Tavares P. Oral myofunctional therapy applied on two cases of severe obstructive sleep apnea. Intl Arch Otorhinolaryngol 2007;11:350–4.
19. Brasili L, Martella S, Vitelli O, et al. Myofunctional treatment of sleep disordered breathing in children [Abstract]. Eur Respir Soc Annu Congress 2013;42:992.
20. Cooper A. Orofacial myology and myofunctional therapy for sleep telated breathing disorders. Sleep Med Clin 2010;5:109–13.
21. Das UM, Beena JP. Effectiveness of circumoral muscle exercises in the developing dentofacial morphology in adenotonsillectomized children: an ultrasonographic evaluation. J Indian Soc Pedod Prev Dent 2009;27:94–103.
22. De Dios JA, Brass SD. New and unconventional treatments for obstructive sleep apnea. Neurotherapeutics 2012;9:702–9.
23. de Paula Silva LM, dos Santos Aureliano FT, Rodrigues Motta A. [Speech therapy in the obstructive sleep apnea-hypopnea syndrome: case report]. Revista CEFAC 2007;9:490–6.

24. Diaféria G, Truksinas E, Haddad FLM, et al. Phonoaudiological assessment of patients with obstructive sleep apnea. Sleep Sci 2011;4:1–7.
25. Guimaraes KC, Drager LF, Marcondes BF, Lorenzi Filho G. Treatment of obstructive sleep apnea with oro-pharyngeal exercises: a randomized study [abstract]. Am J Respir Crit Care Med 2007:A755.
26. Guimaraes KC, Protetti HM. The phonoaudiological work at obstructive sleep apnea. Sleep 2003;26(Abstract Suppl):A209.
27. Huang YS, Guilleminault C. Pediatric obstructive sleep apnea and the critical role of oral-facial growth: evidences. Front Neurol 2012;3:184.
28. Khaleghipour S, Masjedi M, Kelishadi R. The effect of breathing exercises on the nocturnal enuresis in the children with the sleep-disordered breathing. Iran Red Crescent Med J 2013;15:e8986.
29. Kronbauer KF, Trezza PM, Gomes CF. [Speech therapy proposals to the snoring patient]. Distúrbios da Comunicação 2013;25:119–27.
30. Moeller JL. Orofacial myofunctional therapy: why now? Cranio 2012;30:235–6.
31. Sauer C, Schluter B, Hinz R, Gesch D. Childhood obstructive sleep apnea syndrome: an interdisciplinary approach A prospective epidemiological study of 4,318 five-and-a-half-year-old children. J Orofac Orthop 2012;73:342–58.
32. Valbuza JS, de Oliveira MM, Conti CF, Prado LB, de Carvalho LB, do Prado GF. Methods for increasing upper airway muscle tonus in treating obstructive sleep apnea: systematic review. Sleep Breath 2010;14:299–305.
33. Valbuza JS, Oliveira MM, Conti CF, Prado LB, Carvalho LB, Prado GF. Methods to increase muscle tonus of upper airway to treat snoring: systematic review. Arq Neuropsiquiatr 2008;66:773–6.
34. Verma SK, Maheshwari S, Sharma NK, Prabhat KC. Role of oral health professional in pediatric obstructive sleep apnea. Natl J Maxillofac Surg 2010;1:35–40.
35. Villa MP, Brasili L, Ferretti A, et al. Oropharyngeal exercises to reduce symptoms of OSA after AT. Sleep Breath 2014.
36. Moeller JL, Paskay LC, Gelb ML. Myofunctional therapy: a novel treatment of pediatric sleep-disordered breathing. Sleep Med Clin 2014;9:235–43.
37. Kumar TV, Kuriakose S. Ultrasonographic evaluation of effectiveness of circumoral muscle exercises in adenotonsillectomized children. J Clin Pediatr Dent 2004;29:49–55.
38. Johns MW. A new method for measuring daytime sleepiness: the Epworth sleepiness scale. Sleep 1991;14:540–5.
39. Johns M, Hocking B. Daytime sleepiness and sleep habits of Australian workers. Sleep 1997;20:844–9.

Table S1—General characteristics of included patients and quality criteria of included studies.

General Characteristics							Quality Assessment of Included Studies[a]							
Authors, Year	**Site**	**Design**	**N**	**Follow-up**	**BMI**	**Outcomes Analyzed**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**
Pediatric Studies														
Villa et al., 2014	Italy	RT	14	2 mo	21.6	AHI, O_2 sat	No	Yes	Yes	Yes	Yes	No	Yes	Yes
Guilleminault et al., 2013	USA	RCS	11	4 y	–	AHI, O_2 sat	Yes	Yes	Yes	Yes	No	No	Yes	Yes
Adult Studies														
Suzuki et al., 2013	Japan	PCS	6	2 mo	23.8	AHI, O_2 sat	No	Yes	Yes	Yes	Yes	No	Yes	No
Kronbauer et al., 2013	Brazil	PCS	8	2.5 mo	–	ESS, physical measurements	No	Yes	No	Yes	Yes	No	Yes	No
Diaferia et al., 2013	Brazil	RCT	27	3 mo	25.0	AHI, AI, ESS, O_2 sat	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Baz et al., 2012	Egypt	PCS	30	3 mo	33.6	AHI, ESS, O_2 sat, snoring	No	Yes	Yes	Yes	Yes	No	Yes	No
Guimaraes et al., 2009	Brazil	RCT	16	3 mo	29.6	AHI, AI, ESS, O_2 sat, snoring	No	Yes	Yes	Yes	Yes	No	Yes	Yes
de Paula Silva et al., 2007	Brazil	RCR	1	–	23.3	AHI, O_2 sat, sleepiness, snoring	NA	NA	NA	NA	NA	NA	NA	NA
Berreto et al., 2007	Brazil	RCS	2	4 mo	24.2	AHI, ESS, O_2 sat, snoring	No	No	No	Yes	No	No	No	No
Guimaraes et al., 2003	Brazil	ABS	10	–	–	AHI, ESS	NA	NA	NA	NA	NA	NA	NA	NA
Guimaraes et al., 1999	Brazil	RCS	20	6 mo	–	AHI, sleepiness	No	Yes	No	Yes	Yes	No	Yes	No

[a]Quality assessment of cases series studies checklist from National Institute for Health and Clinical Excellence (NICE): (1) Case series collected in more than one center,i.e.,multicenter study? (2) Is the hypothesis/aim/objective of the study clearly described? (3) Are the inclusion and exclusion criteria (case definition) clearly reported? (4) Is there a clear definition of the outcomes reported? (5) Were data collected prospectively? (6) Is there an explicit statement that patients were recruited consecutively? (7) Are the main findings of the study clearly described? (8) Are outcomes stratified? (e.g., by disease stage,abnormal test results, patient characteristics)? –, not reported; AI, apnea index; AHI, apnea-hypopnea index; ESS, Epworth Sleepiness Scale; mo, months; N, number of patients with intervention; NA, not applicable; O_2 sat, oxygen saturation; PCS, prospective case series; RCR, retrospective case report; RCS, retrospective case series; RCT, randomized controlled trial; RT, randomized trial; y, years.