

MYOFUNCTIONAL THERAPY TO TREAT OSA: REVIEW AND META-ANALYSIS

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 \pm standard deviation ($M \pm SD$) of $24.5 \pm 14.3/h$ to $12.3 \pm 11.8/h$, mean difference (MD) -14.26 [95% confidence interval (CI) $-20.98, -7.54$], $P < 0.0001$. Lowest oxygen saturations improved from $83.9 \pm 6.0\%$ to $86.6 \pm 7.3\%$, MD 4.19 (95% CI $1.85, 6.54$), $P = 0.0005$. Polysomnography snoring decreased from $14.05 \pm 4.89\%$ to $3.87 \pm 4.12\%$ 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 $4.87 \pm 3.0/h$ to $1.84 \pm 3.2/h$, $P = 0.004$. 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 $0.5 \pm 0.4/h$), whereas 13 controls had recurrent OSA (AHI $5.3 \pm 1.5/h$) 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

Citation: Camacho M, Certal V, Abdullatif J, Zaghi S, Ruoff CM, Capasso R, Kushida CA. Myofunctional therapy to treat obstructive sleep apnea: a systematic review and meta-analysis. *SLEEP* 2015;38(5):669–675.

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

Submitted for publication July, 2014

Submitted in final revised form August, 2014

Accepted for publication August, 2014

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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 study³⁵ 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/m²) 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 \pm standard deviation (M \pm 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 I² 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 I² 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 $83.9 \pm 6.0\%$ to $86.6 \pm 7.3\%$, 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 I² statistic (59%) and the Q statistic (value of 0.05)

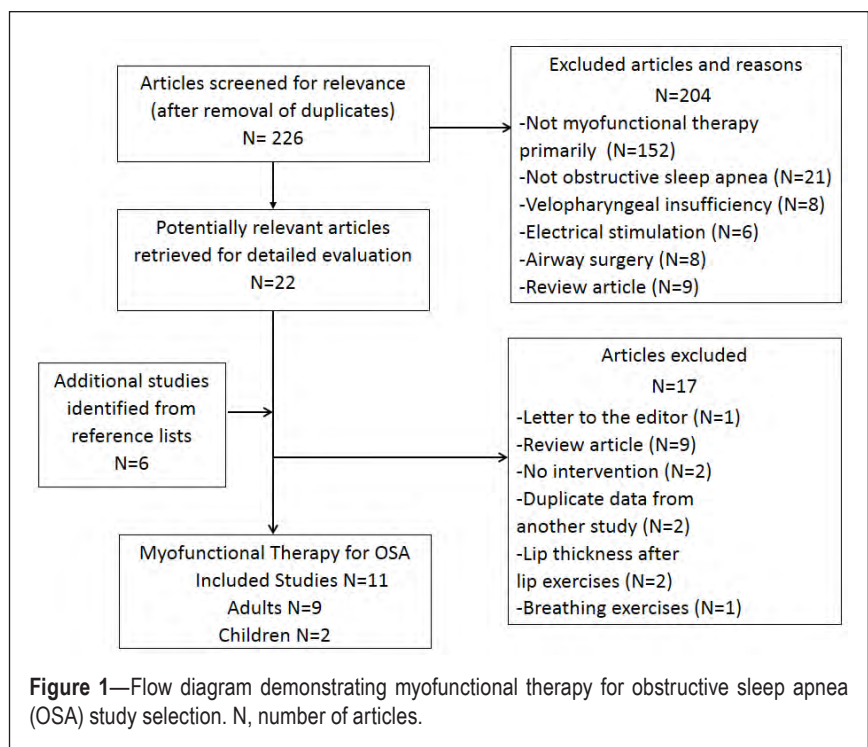


Figure 1—Flow diagram demonstrating myofunctional therapy for obstructive sleep apnea (OSA) study selection. N, number of articles.

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

Authors, Year	Study Design	N	Age (years)	BMI (kg/m ²)	AHI (events/h)		low O ₂ (%)		ESS	
					Pre-MT	Post-MT	Pre-MT	Post-MT	Pre-MT	Post-MT
Suzuki et al., 2013*	PCS	6	22.0 \pm 0.5	23.8 \pm 1.8	15.1 \pm 3.4	9.2 \pm 1.5	90.0 \pm 2.9	96.8 \pm 0.8	—	—
Kronbauer et al., 2013	PCS	8	(40–65)	—	—	—	—	—	11.75	4.25
Diaferia et al., 2013	RCT	27	45.2 \pm 13.0	25.0 \pm 7.4	28.0 \pm 22.7	13.9 \pm 18.5	83.7 \pm 7.7	84.9 \pm 8.8	13.7 \pm 3.2	7.5 \pm 3.7
Baz et al., 2012	PCS	30	44.1 \pm 7.5	33.6 \pm 2.0	22.3 \pm 4.5	11.5 \pm 5.4	84 \pm 4	87 \pm 5	16.4 \pm 2.0	9.3 \pm 2.9
Guimaraes et al., 2009	RCT	16	51.5 \pm 6.8	29.6 \pm 3.8	22.4 \pm 4.8	13.7 \pm 8.5	83 \pm 6	85 \pm 7	14 \pm 5	8 \pm 6
de Paula Silva et al., 2007	RCR	1	60	23.3	44	3	83	92	—	—
Berreto et al., 2007	RCS	2	46 \pm 12.7	24.2 \pm 2.9	44.5 \pm 5.7	6.0 \pm 3.7	78 \pm 1.4	85 \pm 2.8	12.5 \pm 0.7	4.5 \pm 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 \pm 11.6	28.9 \pm 6.2	24.5 \pm 14.3	12.3 \pm 11.8	83.9 \pm 6.0	86.6 \pm 7.3	14.8 \pm 3.5	8.2 \pm 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/m², kilograms per meter squared; low O₂, 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.

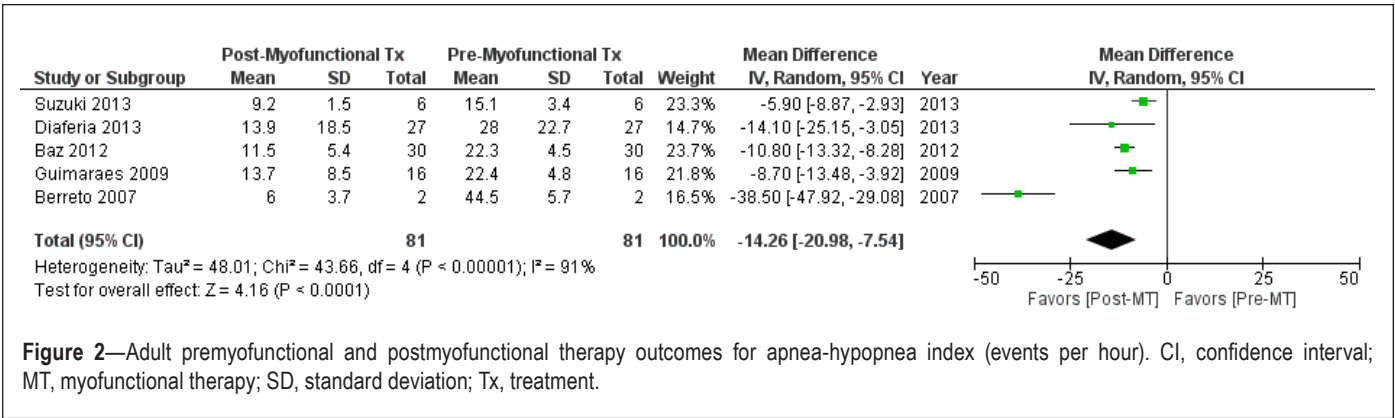


Figure 2—Adult premyofunctional and postmyofunctional therapy outcomes for apnea-hypopnea index (events per hour). CI, confidence interval; MT, myofunctional therapy; SD, standard deviation; Tx, treatment.

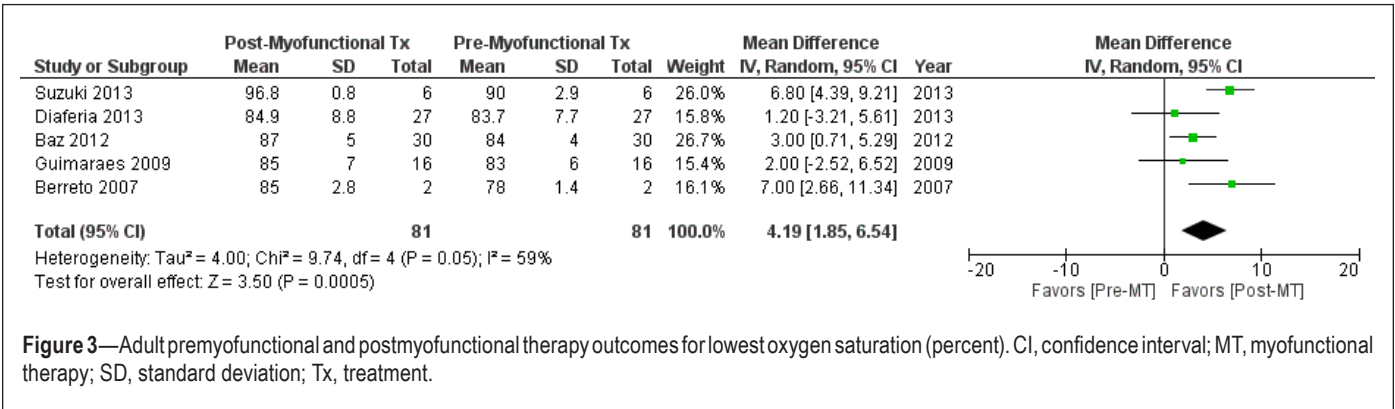


Figure 3—Adult premyofunctional and postmyofunctional therapy outcomes for lowest oxygen saturation (percent). CI, confidence interval; MT, myofunctional therapy; SD, standard deviation; Tx, treatment.

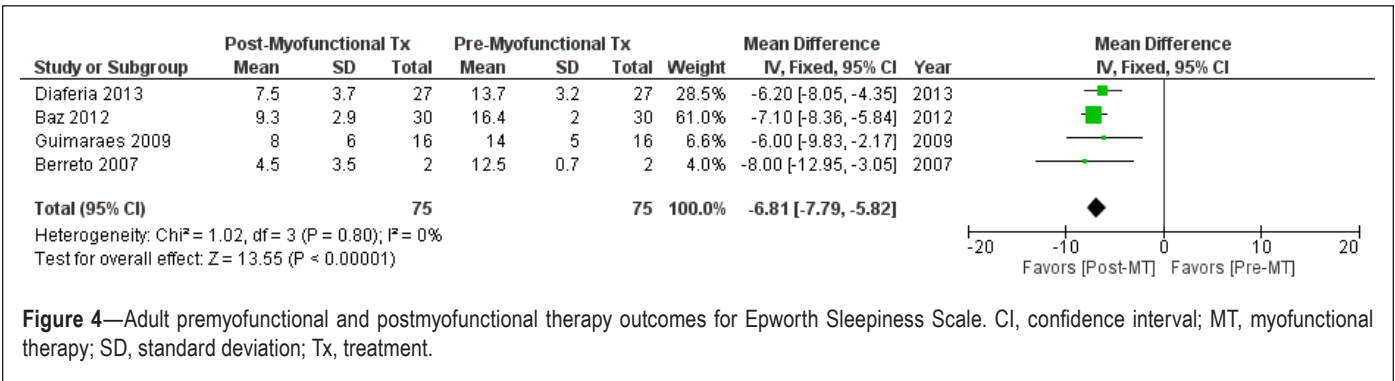


Figure 4—Adult premyofunctional and postmyofunctional therapy outcomes for Epworth Sleepiness Scale. CI, confidence interval; MT, myofunctional therapy; SD, standard deviation; Tx, treatment.

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 I² 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 I² 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 $M \pm SD$ reduced from $4.87 \pm 3.0/h$ to $1.84 \pm 3.2/h$, $P = 0.004$ (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 $5.3 \pm 1.5/h$).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 $24.5 \pm 14.3/h$ to $12.3 \pm 11.8/h$, MD of -14.26 [95% CI $-20.98, -7.54$] ($P < 0.0001$). For pediatric patients, the pre- and post-MT $M \pm SD$ for AHI decreased from $4.87 \pm 3.0/h$ to $1.84 \pm 3.2/h$, $P = 0.004$. Additionally, the study by Guilleminault et al.¹⁰ reported that 11 children remained cured of OSA (AHI of $0.5 \pm 0.4/h$) 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 ($14.05 \pm 4.89\%$ to $3.87 \pm 4.12\%$, 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 $M \pm SD$ s 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 Guimaraes⁹ 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 mo¹² 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. Guimaraes⁹ 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 outcomes⁷; 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 $M \pm SD$ 29.6 \pm 3.8) and obese patients (BMI $M \pm SD$ 33.6 \pm 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.

DISCLOSURE STATEMENT

This was not an industry supported study. Dr. Kushida has received research support from Aerial BioPharma, Pacific Medico Co., Ltd., Resmed, Apnex Medical, Impax Laboratories, Inc., and Cephalon; has consulted for Apnex, Seven Dreamers Laboratories, Noven Pharmaceuticals, UCB, Philips-Respiroics, Zephyr; and has received royalties from Philips Respiroics. The other authors have indicated no financial conflicts

of interest. The work was performed at Stanford Hospital and Clinics, Stanford, CA. The views herein are the private views of the authors and do not reflect the official views of the Department of the Army or the Department of Defense.

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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

^aQuality 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; O₂ sat, oxygen saturation; PCS, prospective case series; RCR, retrospective case report; RCS, retrospective case series; RCT, randomized controlled trial; RT, randomized trial; y, years.



Oropharyngeal and tongue exercises (myofunctional therapy) for snoring: a systematic review and meta-analysis

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Received: 26 November 2017 / Accepted: 14 December 2017

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Abstract

Purpose Oropharyngeal and tongue exercises (myofunctional therapy) have been shown to improve obstructive sleep apnea. However, to our knowledge, a systematic review has not been performed for snoring. The study objective is to perform a systematic review, with a meta-analysis, dedicated to snoring outcomes after myofunctional therapy.

Methods PubMed/MEDLINE and three other databases were searched through November 25, 2017. Two authors independently searched the literature. Eligibility (1) patients: children or adults with snoring, (2) intervention: oropharyngeal and/or tongue exercises, (3) comparison: pre and post-treatment data for snoring, (4) outcomes: snoring frequency and snoring intensity, (5) study design: publications of all study designs.

Results A total of 483 articles were screened, 56 were downloaded in their full text form, and nine studies reported outcomes related to snoring. There were a total of 211 patients (all adults) in these studies. The snoring intensity was reduced by 51% in 80 patients from pre-therapy to post-therapy visual analog scale values of 8.2 ± 2.1 (95% CI 7.7, 8.7) to 4.0 ± 3.7 (95% CI 3.2, 4.8). Berlin questionnaire snoring intensity reduced by 36% in 34 patients from 2.5 ± 1.0 (95% CI 2.2, 2.8) to 1.6 ± 0.8 (95% CI 1.3, 1.9). Finally, time spent snoring during sleep was reduced by 31% in 60 patients from $26.3 \pm 18.7\%$ (95% CI 21.6, 31.0) to $18.1 \pm 20.5\%$ (95% CI 12.9, 23.3) of total sleep time.

Conclusions This systematic review demonstrated that myofunctional therapy has reduced snoring in adults based on both subjective questionnaires and objective sleep studies.

Keywords Snoring · Myofunctional therapy · Systematic review · Meta-analysis

The views expressed in this abstract/manuscript are those of the authors and do not reflect the official policy or position of the Department of the Army, Department of Defense, or the US Government.

Electronic supplementary material The online version of this article (<https://doi.org/10.1007/s00405-017-4848-5>) contains supplementary material, which is available to authorized users.

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Introduction

There have been several treatments developed over the years to treat snoring and obstructive sleep apnea (OSA) [1–4]. Of the current techniques to treat snoring and OSA, many of them are invasive and involve either performing surgery or wearing a device during sleep [5–7]. A technique that can serve either as a primary treatment or as an adjunct treatment to treat primary snoring that does not require either surgery or wearing a device would be beneficial. Tongue exercises and oropharyngeal exercises (myofunctional therapy) have improved OSA in children and adults [8]. In a previous meta-analysis evaluating myofunctional therapy, apnea-hypopnea index was reduced by 50% in adults and 62% in children [8]. The sub-analysis, evaluating patients with sleep study snoring, demonstrated a significant reduction from $14.05 \pm 4.89\%$ to $3.87 \pm 4.12\%$ of total sleep time, p value < 0.001 [8].

Since the publication of the meta-analysis for OSA, there have been several studies evaluating oropharyngeal exercises and tongue exercises and their outcomes for snoring; however, to our knowledge, there has been no systematic review or meta-analysis evaluating the effect on snoring. To provide the most up-to-date information, a systematic review would be required. Therefore, the objective of this study was to perform a systematic review for snoring, specifically using the PICOS acronym, as follows: (1) *Patients (P)* adults or children who snore; (2) *Intervention (I)* oropharyngeal exercises and/or tongue exercises; (3) *Comparison (C)* data pre and post-exercises; (4) *Outcomes (O)* snoring frequency, snoring index, percentage of night spent snoring, visual analog scale (VAS), and Likert scales; (5) *Study design (S)* any study type or design. After obtaining the studies, the pre- and post-oropharyngeal exercises and tongue exercises snoring data were analyzed.

Methods

The preferred reporting items for systematic reviews and meta-analysis (PRISMA) statement were reviewed and used as a guide during this study [9].

Protocol

Our Institutional Department of Clinical Investigation was contacted, and a protocol was submitted and was approved. For this type of study, formal consent is not required.

Eligibility criteria

The inclusion criteria for this review: (1) studies with adult or pediatric patients who were treated with oropharyngeal exercises and tongue exercises as the sole intervention and (2) the publication provided both pre- and post-oropharyngeal exercises and tongue exercises quantitative outcomes for snoring. Exclusion criteria: studies with additional treatments performed, studies using devices, and studies without data for myofunctional therapy alone.

Information sources

We searched PubMed/MEDLINE, Google Scholar, The Cochrane Library and Cumulative Index to Nursing and Allied Health (CINAHL).

Search

Authors M. C. and M. W. N searched through May 8, 2017 initially, and provided additional updating through November 25, 2017. An example of a search strategy is the one

used for PubMed/MEDLINE: [(Snoring OR Sleep) AND (“tongue exercise” OR “tongue exercises” OR “orofacial” OR “myotherapy” OR “speech therapy” OR “oropharyngeal exercises” OR “myofascial reeducation” OR “myofunctional therapy” OR “upper airway exercises” OR (“Myofunctional Therapy”[MeSH]))]. For the remaining databases, we applied very similar keywords and terms, just tailored to the specific databases.

Authors extracted the snoring data from the studies meeting the predefined selection criteria. If a study did not provide the information necessary to include it in the review, then the study authors were emailed at least twice in an attempt to obtain the data.

Risk of bias and heterogeneity

If there are sufficient summary measures provided, then an analysis for bias and heterogeneity would be performed using REVMAN.

Summary measures

Study measures collected include the means, standard deviations (SD), medians, and other summary measures provided by the individual studies.

Results

A total of 483 articles were screened, 56 were downloaded in their entirety, and nine studies [10–18] with 211 patients met the inclusion criteria, see Supplementary Fig. 1. The studies provided data for snoring frequency, snoring intensity, snoring severity, and bedpartner visual analog scale scores, see Table 1. The studies that used Berlin questionnaire and values for snoring frequency were rated as follows: 0 = never, 1 = 1–2 times a month, 2 = 1–2 times a week, 3 = 3–4 times a week, and 4 = every day [19]. Values for snoring intensity were 0 = no snoring, 1 = similar to breathing, 2 = as loud as talking, 3 = louder than talking, and 4 = very loud, and can be heard in adjacent rooms [19].

For the 211 patients who performed myofunctional therapy, the mean snoring frequency and snoring intensity were reduced, see Table 2. In 80 patients, the snoring intensity reduced by 51%, from pre-therapy to post-therapy using the VAS values [from 8.2 ± 2.1 (95% CI 7.7, 8.7) to 4.0 ± 3.7 (95% CI 3.2, 4.8)]. A sub-analysis was performed for VAS using random effects modeling, which demonstrated a mean difference of -3.67 [95% CI $-4.44, -2.90$], overall effect $Z=9.34$, p value <0.00001 , Q statistic p value = 0.64, and $I^2=0\%$ (Fig. 1). The VAS standardized mean difference was -1.46 (95% CI $-1.81, -1.11$), overall effect $Z=8.15$,

Table 1 General characteristics and quality criteria of included studies

Author, year, <i>N</i>	General characteristics			Quality assessment of included studies								
	Country	Design	Snoring data	1	2	3	4	5	6	7	8	
Diaferia et al. 2016, <i>N</i> =27	Brazil	RCT	SF, SI, (VAS)	N	Y	Y	Y	Y	Y	Y	Y	Y
Mohamed et al. 2016, <i>N</i> =30	Egypt	PCS	SF, SI	N	Y	Y	Y	Y	N	Y	N	N
Verma et al. 2016, <i>N</i> =20	India	PCS	SI	N	N	Y	Y	Y	N	Y	N	N
Ieto et al. 2015, <i>N</i> =19	Brazil	RCT	SF, SI (VAS)	N	Y	Y	Y	Y	N	Y	Y	Y
Kayamori et al. 2015, <i>N</i> =30	Brazil	RCT	SF, SI	N	Y	Y	Y	Y	N	Y	Y	Y
Nemati et al. 2015, <i>N</i> =53	Iran	PCS	SS, VAS	N	N	Y	Y	N	N	Y	N	N
Baz et al. 2012, <i>N</i> =30	Egypt	PCS	SF, SI	N	Y	Y	Y	Y	N	Y	N	N
Guimaraes et al. 2009, <i>N</i> =16	Brazil	RCT	SF, SI	N	Y	Y	Y	Y	N	Y	Y	Y
Berreto et al. 2007, <i>N</i> =2	Brazil	RCS	SS	N	N	N	Y	N	N	N	N	N

Columns: (1) case series collected in more than one center, i.e. multi-center 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 abnormal results, disease stage, and patient characteristics)?

PCSprospective case series, RCSretrospective case series, RCTrandomized control trial, SFsnoring frequency, SIsnoring intensity, SSsnoring severity, VASvisual analog scale

Table 2 Demographic and snoring data before and after oropharyngeal exercises and tongue exercises

Study, authors, year	<i>N</i>	Age	BMI	Pre-SF	Post-SF	Pre-SI	Post-SI	% Change SI
Diaferia et al. 2016	27	45 ± 13	25.0 ± 7.4	8.5 ± 2.3 ^{‡V} [7.6–9.4]	4.9 ± 3.2 ^{‡V} [3.7–6.1]	7.7 ± 2.3 ^{‡V} [6.8–8.6]	4.3 ± 2.8 ^{‡V} [3.2–5.4]	–44.2%
Mohamed et al. 2016	30	46.9 ± 6.4	27.9 ± 2.0	464 ± 168 [401–527] ^{‡SN}	396 ± 172 [331–460] ^{‡SN}	38.5 ± 19.5 ^{‡P} [31.5–45.5]	32.3 ± 20.6 ^{‡P} [24.9–39.7]	–16.2%
Verma et al. 2016	20	41 ± 11	25.6 ± 3.1	–	–	2.8 ± 0.5 ^{‡B} [2.6–3.0]	1.7 ± 0.6 ^{‡B} [1.4–2.0]	–39.3%
Ieto et al. 2015	19	48 ± 14	28.1 ± 2.7	4 (3–4) ^{‡B}	2 (1.5–3) ^{‡B}	4 (2.5–4) ^{‡B}	1 (1–2) ^{‡B}	–75%
Kayamori et al. 2015	14	42 ± 13	28.9 ± 4.3	2.7 ± 1.4 ^{‡B} [2.0–3.4]	2.6 ± 1.3 ^{‡B} [1.9–3.3]	2.0 ± 1.4 ^{‡B} [1.3–2.7]	1.5 ± 1.0 ^{‡B} [1.0–2.0]	–25%
Nemati et al. 2015	53	45 ± 10	26.5 ± 5.2	91%	36%	8.5 ± 1.9 ^{‡V} [8.0–9.0]	4.7 ± 2.9 ^{‡V} [3.9–5.5]	–44.7%
Baz et al. 2012	30	44 ± 8	33.6 ± 2.0	100%	53.3%	14.1 ± 4.9 ^{‡P} [12.3–15.9]	3.9 ± 4.1 ^{‡P} [2.4–5.4]	–72.3%
Guimaraes et al. 2009	16	52 ± 7	29.6 ± 3.8	4 (4–4) ^{‡B}	3 (1.5–3.5) ^{‡B}	3 (3–4) ^{‡B}	1 (1–2) ^{‡B}	–66.6%
Berreto et al. 2007	2	46 ± 13	24.2 ± 2.9	–	–	3 ± 0 ^{‡G} [3–3]	2 ± 0 ^{‡G} [2–2]	33.3%

BMIbody mass index, *N*number of patients, SFsnoring frequency, SIsnoring intensity, – not reported

[] Denotes lower and upper 95% confidence intervals

^{‡B}Berlin score, 0–10

^{‡V}Visual analog scale, 0–10.0

^{‡SN}Snores per hour

^{‡G}Grading scale, 0–4

^{‡P}Percent of night based on sleep study

p value < 0.00001, *Q* statistic *p* value = 0.54, and *I*² = 0% (Fig. 1).

In studies that used the Berlin questionnaire, snoring intensity reduced in 34 patients from 2.5 ± 1.0 (95% CI 2.16, 2.84) to 1.6 ± 0.8 (95% CI 1.33, 1.87). A sub-analysis

was performed for Berlin scores for snoring using random effects modeling, which demonstrated a mean difference of –0.95 (95% CI –1.46, –0.44), overall effect *Z* = 3.67, *p* value = 0.0002, *Q* statistic *p* value = 0.22, and *I*² = 33% (Fig. 2). The Berlin scores for snoring using standardized

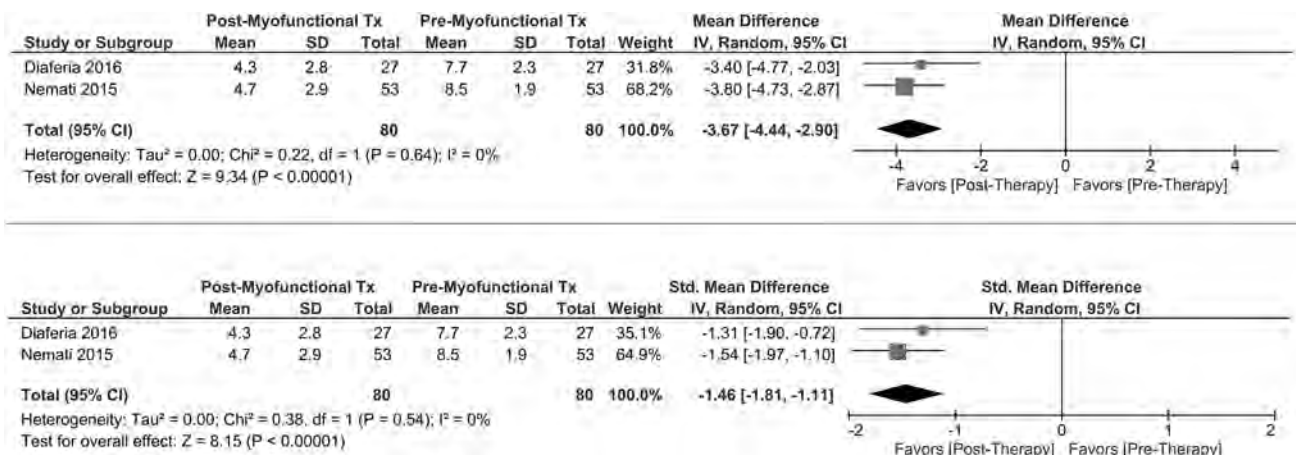


Fig. 1 Pre- and post-myofunctional therapy visual analog scale for snoring intensity. Mean difference (top) and standardized mean difference (bottom)

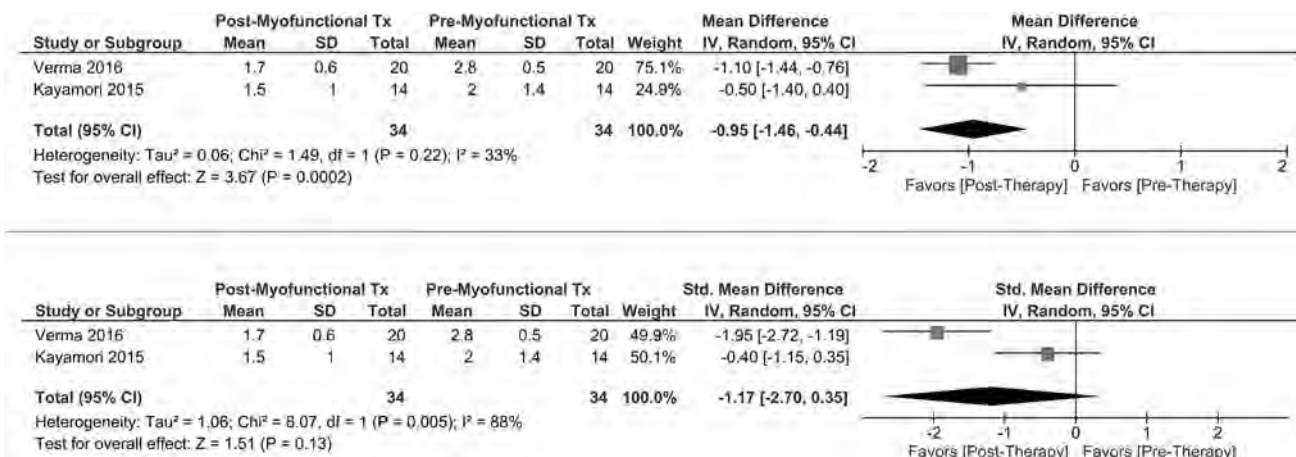


Fig. 2 Pre- and post-myofunctional therapy Berlin score for snoring intensity. Mean difference (top) and standardized mean difference (bottom)

mean difference were -1.17 (95% CI $-2.70, 0.35$), overall effect $Z=1.51$ p value = 0.13, Q statistic p value = 0.005, and $I^2=88\%$ (Fig. 2).

Time spent snoring during sleep was reduced by 31.2% in 60 patients from $26.3 \pm 18.7\%$ (95% CI 21.6, 31.0) to $18.1 \pm 20.5\%$ (95% CI 12.9, 23.3) of total sleep time. A sub-analysis was performed for percentage of time spent snoring with random effects modeling, demonstrating a mean difference of -10.01 percent of the night (95% CI $-12.24, -7.78$), overall effect $Z=8.79$, p value < 0.0001, Q statistic p value = 0.45, and $I^2=0\%$ (Fig. 3). The percentage of time spent snoring's standardized mean difference was -1.26 (95% CI $-3.14, 0.63$) (large effect using Cohen's guidelines), overall effect $Z=1.31$ p value = 0.19, Q statistic p value < 0.00001, and $I^2=95\%$.

Overall, the exercises described were generally performed for 3 months and consisted of four main locations, the soft palate, the tongue, facial exercises, pharyngeal exercises,

jaw exercises, and stomatognathic exercises [10–18]. Soft palate exercises generally consisted of saying vowels, which recruits the palatoglossus, palatopharyngeus, tensor veli palatini, levator veli palatini, and the uvula [12]. Tongue exercises generally consisted of moving the tongue in different directions with or without sticking the tongue out, pressing against bony and soft tissue structures within the oral cavity, sucking the tongue against the palate, and other tongue movements with or without resistance [10–18]. Facial exercises generally involve recruitment of the buccinator muscles by placing a finger into the oral cavity and pressing in an outward direction and puckering, closing or moving the lips [10–18]. Jaw exercises involve opening/closing/exercising the jaw. Pharyngeal exercises can involve swallowing exercises. Finally, stomatognathic functional exercises can involve sucking through a narrow straw, inflating balloons and swallowing and chewing exercises.

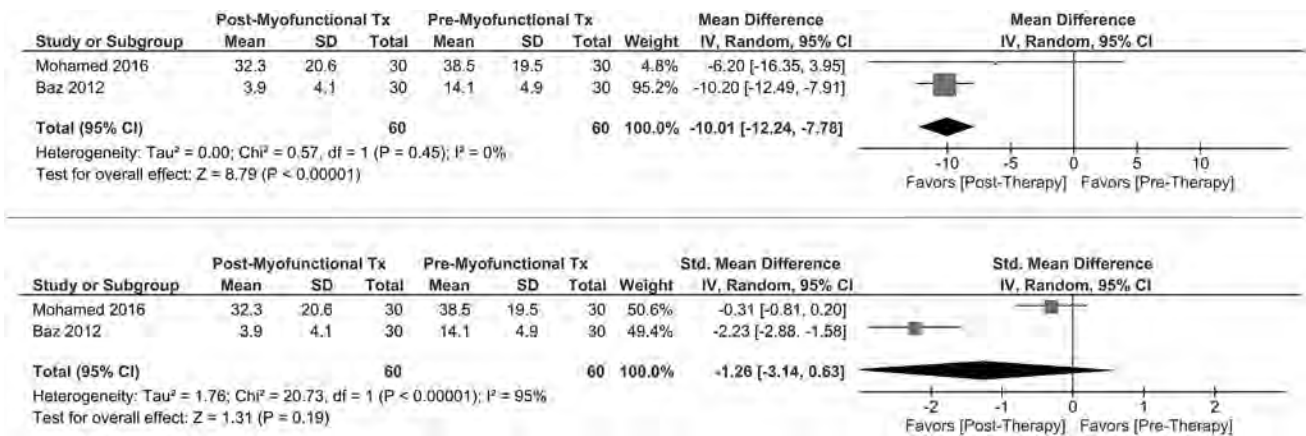


Fig. 3 Pre- and post-myofunctional therapy percentage of time spent snoring during the sleep study. Mean difference (top) and standardized mean difference (bottom)

Individual studies

Diaferia et al. [12] evaluated 100 patients who were randomized into various treatments and 27 were placed into the myofunctional therapy treatment arm. The myofunctional therapy consisted of tongue, soft palate, stomatognathic function, and facial exercises [12]. The patients performed the exercises three times daily, for 20 min sessions, a total of 3 months [12]. The snoring frequency using the visual analog scale was 8.5 ± 2.3 and 4.9 ± 3.2 (42% reduction) before and after myofunctional therapy [12]. The snoring intensity reduced from 7.7 ± 2.3 to 4.3 ± 2.8 before and after treatment, corresponding to a 44% reduction) [12].

Mohamed et al. [18] treated 30 patients with OSA by having them perform oropharyngeal exercises (soft palate, tongue, facial muscles, and stomatognathic function exercises) [18]. Exercises were performed for at least 10 min, three to five times a day for 3 months. The patients were divided into two groups (Group 1 with moderate OSA and Group 2 with severe OSA) [18]. Snoring index in patients with moderate OSA reduced by 24%, and the percent time spent snoring during the sleep study decreased by 37%. However, in patients with severe OSA, the snoring index only reduced by 10%, and the percent time spent snoring during the sleep study only reduced by 9%.

Verma et al. [17] evaluated 20 patients who were treated with myofunctional therapy. The exercises were performed five times daily, for 3 months [17]. The exercises performed included tongue exercises, jaw exercises, lip exercises, and soft palate exercises [17]. The researchers used the Berlin scoring for snoring. The snoring intensity was reduced from 2.8 ± 0.5 before myofunctional therapy down to 1.7 ± 0.6 after myofunctional therapy (a 39% reduction) [17].

Ieto et al. [14] treated nineteen patients with myofunctional therapy to include tongue exercises, palate exercises,

facial exercises, and chewing/swallowing exercises. The patients performed the myofunctional therapy exercises for approximately 8 min daily for 3 months [14]. The researchers used the Berlin scoring. The median values for snoring frequency were reported and were 4 (3–4) before myofunctional therapy and 2 (1.5–3) after myofunctional therapy [14]. The snoring intensity reduced from 4 (2.5–4) before treatment, down to 1 (1–2) after treatment [14].

Kayamori and Filho [15] had 14 patients who underwent myofunctional therapy and had data that could be analyzed. The exercises were performed three times a day for 3 months [15]. Exercises included tongue exercises, soft palate exercises, facial exercises, and chewing/swallowing exercises [15]. The researchers used the Berlin scoring. The authors found that the snoring frequency did not change significantly 2.7 ± 1.4 to 2.6 ± 1.3 (4% reduction); however, the snoring intensity did decrease from 2.0 ± 1.4 to 1.5 ± 1.0 (25% reduction) [15].

Nemati et al. [16] reported treating 53 patients with primary snoring with myofunctional therapy for 30 min sessions, 5 days a week for 3 months. Patients performed soft palate exercises, tongue exercises, and facial exercises [16]. The researchers used the Lim and Curry snoring scale score (SSS) [20], frequency of snoring (every night, most nights, some nights, and seldom/never), the duration of snoring (all night long, most hours of the night, or some hours of the night), and the visual analog scale (0–10) [16]. The snoring severity scale demonstrated a reduction in snoring from 7.0 ± 1.7 to 3.1 ± 2.7 (56% reduction) [16]. The frequency of snoring based on the percentage of patients who snored every night or most nights was reduced from 91 to 36% [16]. The visual analog scale demonstrated an improvement in snoring from 8.5 ± 1.9 to 4.7 ± 2.9 (45% reduction) [16].

Baz et al. [10] evaluated 30 patients based on symptoms and a sleep study. The patients performed exercises for at

least 10 min, 3–5 times daily for 3 months [10]. The myofunctional therapy included tongue exercises, the soft palate exercises, and pharyngeal exercises. Before myofunctional therapy, 100% of patients snored and afterwards 53% snored [10]. The sleep study demonstrated a reduction in the total time spent snoring from $14.1 \pm 4.9\%$ down to $3.9 \pm 4.1\%$, which is a 72% reduction [10].

Guimaraes et al. [13] reported outcomes for 16 patients who were treated with myofunctional therapy for 3 months. Exercises performed included the tongue exercises, soft palate exercises, facial exercises, and stomatognathic function exercises [13]. The snoring frequency and intensity were obtained using the Berlin questionnaire. The median values for snoring frequency reduced from 4 (4–4) down to 3 (1.5–3.5) [13]. The median values for snoring intensity reduced from 3 (3–4) down to 1 (1–2), a 67% reduction [13].

Berreto et al. [11] had two patients who performed myofunctional therapy for 16 weeks. Exercises included tongue exercises, facial exercises, soft palate exercises, pharyngeal exercises, jaw exercises, and stomatognathic function exercises. Snoring was grades 0–4, where 0 = no snoring, 1 = heavy breathing, 2 = light snoring, 3 = snoring that disturbs the bedpartner, and 4 = snoring that disturbs the family [11]. The snore score decreased from 3 to 2 for both patients, corresponding to a 33% reduction [11].

Discussion

There are three main findings from this systematic review. First, the systematic review has demonstrated an improvement in snoring by approximately 50% after myofunctional therapy. An improvement is seen in all the study measures (Berlin questionnaires, VAS, and snoring during the sleep study). The studies have all been in adult patients thus far, and to our knowledge, a pediatric study has not reported outcomes for snoring. Interestingly, the 50% improvement in snoring seen in adults is consistent with the improvement seen in OSA (also 50%) in the meta-analysis performed for myofunctional therapy and OSA [8]. In addition, there was objective improvement in snoring based on polysomnography, with a 31% improvement in the percentage of time spent snoring.

Second, pediatric studies are lacking. Although there are no pediatric studies evaluating snoring, there was a significant improvement in pediatric OSA after myofunctional therapy in the previous meta-analysis [8]. Therefore, it is likely that the improvement in snoring would have also been noted in children; however, we cannot generalize, since there were no studies identified. Anecdotally, a few of the authors' (MC, CG, and SZ) pediatric patients undergoing myofunctional therapy as adjunct or primary treatment for snoring or OSA have been noted

to have significant decreases in the snoring intensity and frequency. Interestingly, there is debate regarding snoring in pediatric patients: younger children have a greater chance of sleeping closer to their parents, while older pre- and peri-pubertal children usually sleep farther away from where parents sleep; therefore, the parents are more likely to hear younger children. This snoring phenomenon is even more true for pubertal and post-pubertal teenagers: therefore, there is a clear change in the possibility of perception of snoring during childhood and this has been pointed out in different pediatrics studies. In adults, there is a bias on reporting given that snoring complaints are bedpartner driven; therefore, adults who sleep alone generally do not have people complain unless they share a room for some reason. This bedpartner phenomenon presents a risk of bias concerning snoring outcomes, but despite this potential bias, the studies were consistent in their findings of decreased snoring noted after myofunctional therapy.

Third, although there are improvements in snoring, the mechanism of action as to why myofunctional therapy improves snoring are not completely understood. Given that the lips, facial muscles, tongue, soft palate, oral cavity, and pharynx are exercised by the techniques used in the studies in this manuscript, we hypothesize that the training improves both tone and positioning. An analogy could be seen in people who have never lifted weights and want to start weight training; initially, they will not be able to lift as much weight, but after lifting for 3 months, they will have improved strength and tone. It is possible, therefore, that the myofunctional therapy can help improve the tone and strength of the oral cavity, tongue, soft palate, and pharynx analogous to the improvement in strength and tone that is seen with weight training. Friberg et al. demonstrated that heavy snorers have a neuropathy of the soft palate when compared to control patients and there is even more neuropathy in patients with OSA [21]. Engelke et al. explored orofacial training and hypothesized that it promotes a closed oral rest position which can help to keep the tongue in contact with the palate and lead to an intraoral negative pressure which may help stabilize the pharynx into a more open position (and may also reduce the neuromuscular activity necessary to maintain the open airway) [22].

Limitations

As with all systematic reviews, we are limited to the currently published studies. It is possible that authors who have not seen a difference in snoring outcomes for their patients did not submit their findings, or if they did submit their findings, then maybe their study was not accepted secondary to publication bias against negative studies.

Conclusions

This systematic review demonstrated that myofunctional therapy has reduced snoring in adults based on both subjective questionnaires and objective sleep studies. No pediatric studies were identified. Additional research is recommended based on these initial encouraging results.

Funding No funding was received for this research.

Compliance with ethical standards

Conflict of interest All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria, educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge, or beliefs) in the subject matter or materials discussed in this manuscript.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent For this type of study, formal consent is not required. There is no additional need for informed consent as no identifying information is included in this article.

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