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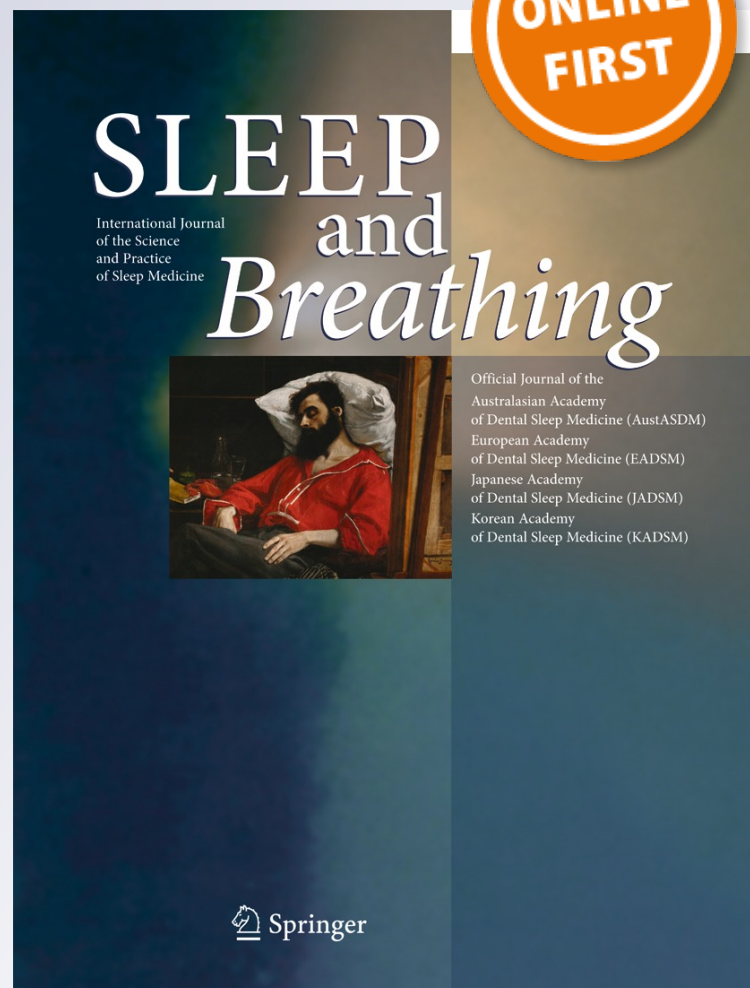
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Myofunctional therapy improves adherence to continuous positive airway pressure treatment

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Abstract

Purpose Few studies have investigated myofunctional therapy in patients with obstructive sleep apnea syndrome (OSAS). The objective of this study was to evaluate the effect of myofunctional therapy on continuous positive airway pressure (CPAP) adherence.

Methods The study was registered at ClinicalTrials.gov (NCT01289405). Male patients with OSAS were randomly divided into four treatment groups: placebo, patients undergoing placebo myofunctional therapy ($N = 24$); myofunctional therapy, undergoing myofunctional therapy ($N = 27$); CPAP, undergoing treatment with CPAP ($N = 27$); and combined, undergoing CPAP therapy and myofunctional therapy ($N = 22$). All patients underwent evaluations before and after 3 months of treatment evaluation and after 3 weeks of washout. Evaluations included Epworth sleepiness scale (ESS), polysomnography, and myofunctional evaluation.

Results The 100 men had a mean age of 48.1 ± 11.2 years, body mass index of 27.4 ± 4.9 kg/m², ESS score of 12.7 ± 3.0 , and apnea-hypopnea index (AHI) of 30.9 ± 20.6 . All treated groups (myofunctional therapy, CPAP, and combined myofunctional therapy with CPAP) showed decreased ESS and snoring, and the myofunctional therapy group maintained

this improvement after the “washout” period. AHI reduction occurred in all treated groups and was more significant in CPAP group. The myofunctional therapy and combined groups showed improvement in tongue and soft palate muscle strength when compared with the placebo group. The association of myofunctional therapy to CPAP (combined group) showed an increased adherence to CPAP compared with the CPAP group.

Conclusions Our results suggest that in patients with OSAS, myofunctional therapy may be considered as an adjuvant treatment and an intervention strategy to support adherence to CPAP.

Keywords Obstructive sleep apnea · Treatment · Myofunctional therapy · Continuous positive airway pressure · Polysomnography

Introduction

Obstructive sleep apnea syndrome (OSAS) is a disease with multifactorial pathways of pathophysiology that involve anatomical and functional pharyngeal changes [1, 2]. Although the treatment of choice is the continuous positive airway pressure (CPAP) device, adherence to this type of therapy has been a limiting factor [3].

Myofunctional therapy may promote increased tongue, pharynx, and soft palate muscle tone, through isometric exercises (working with muscle tension) and isotonic exercises (improving mobility), constituting a possible therapeutic option to OSAS [4, 5]. The myofunctional therapy treats OSAS and could reduce pressure and improve adherence to CPAP use. However, studies that demonstrate the effect of this treatment in OSAS, particularly when associated with CPAP, remain scarce in the literature. One hypothesis is that by

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strengthening the muscles of the pharynx, myofunctional therapy reduces the required level of CPAP and may increase adherence to CPAP treatment. Moreover, myofunctional therapy may be associated with educational support and appropriate guidance to the side effects during treatment with CPAP [6, 7]. Due to the lack of studies in this field of research, the present study evaluated the effect of myofunctional therapy on CPAP adherence.

Methods

Consecutive men, age 25 to 65 years, body mass index (BMI) $<35 \text{ kg/m}^2$, and with OSAS diagnosis confirmed by clinical and polysomnographic criteria [8] were included in this study. Exclusion criteria included the following: female gender since hormonal decline in the menopausal phase could lead to loss of muscle mass, causing a bias in the study; patients who were uncooperative, illiterate, or who had a low education level that prevented the completion of questionnaires and the understanding of the guidelines about the use of CPAP and practice of exercises that have been written to be practiced at home; patients with other sleep disorders or with previous treatment for OSAS (i.e., CPAP, intraoral device, or surgery); patients with serious or decompensated clinical or psychiatric medical illnesses, such as congestive heart failure, cardiomyopathy, chronic obstructive pulmonary disease, chronic active hepatitis, liver cirrhosis with severe symptoms, myasthenia gravis, demyelinating disease, motor neuron disease, depression, schizophrenia, obsessive compulsive disorder, disorder anxiety, bipolar disorder, eating disorder, attention deficit disorder, and hyperactivity; patients who used alcohol, stimulants or sedatives; and patients with grade III or IV palatine tonsils, grade II or III septal deviation, or evident micrognathia.

The study was approved by the research ethics committee of the Universidade Federal de São Paulo—UNIFESP (CEP 2002/08) and registered at [ClinicalTrials.gov](https://www.clinicaltrials.gov) (NCT01289405). All patients gave written informed consent.

Prior to treatment, the patients were divided randomly into four groups: the placebo group, which included patients undergoing placebo myofunctional therapy; the myofunctional therapy group, which included patients undergoing myofunctional therapy; the CPAP group, which included patients undergoing CPAP treatment without myofunctional or placebo interventions; and the combined group, which included patients undergoing both CPAP and myofunctional therapy.

All patients underwent an evaluation protocol three times during the study: before treatment, after 3 months of

treatment, and after a 3-week washout period (Fig. 1). During the clinical assessment, the evaluators were blinded.

Questionnaires

All patients completed the Epworth sleepiness scale (ESS) [9] for assessment of subjective sleepiness; a visual analog scale for subjective evaluation of snoring; and diaries from the CPAP Clinic of the Sleep Institute for subjective adherence, comfort, and satisfaction using the nasal CPAP treatment. A follow-up questionnaire was also completed based on a protocol that approaches physical, psychological, cognitive, and social conditions, relating them to possible sleep disorders. In addition, it evaluated adherence and side effects of CPAP treatment. The questionnaire also included fields to be filled out with detailed data obtained within the equipment.

Myofunctional evaluation

The myofunctional status was evaluated according to protocols established in the literature [10, 11] and included tongue and soft palate strength investigation and the Modified Mallampati Index (MMI).

Polysomnography

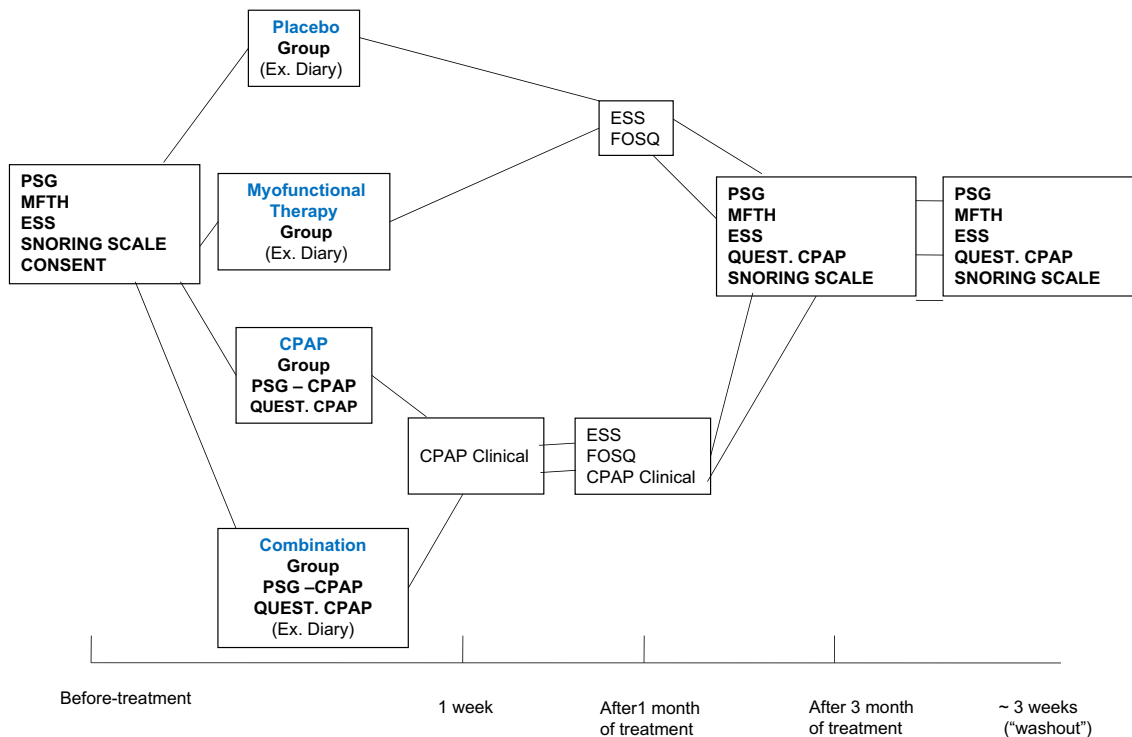
Full night inlab attended polysomnography (PSG) was performed with a computerized system (EMBLA® S7000, Embla Systems, Inc., Broomfield, CO, USA).

Sleep staging was performed according to the criteria proposed by Rechtschaffen and Kales [12], and the arousals followed the criteria of the American Sleep Disorders Association [13]. The respiratory events were analyzed according to the criteria proposed by the American Academy of Sleep Medicine [14]. OSAS severity was defined as follows: mild = apnea-hypopnea index (AHI) from 5 to 14.9 events/h, moderate = AHI from 15 to 29.9/h, and severe = AHI 30 or more events/h [14].

Placebo and myofunctional therapy interventions

Placebo therapy consisted of exercises without therapeutic function (relaxation and stretching of the neck muscles), as described by Cunali and coworkers [15]. Myofunctional therapy consisted of muscular endurance exercises aimed at toning the oropharynx muscle groups; optimizing muscle tension mobility; and adjusting the position of the soft tissues and the suitability of the chewing, sucking, swallowing, and breathing orofacial functions, according to previously standardized protocols [4, 16–22].

Myofunctional therapy includes soft palate, tongue, and facial muscle exercises as well as stomatognathic function exercises, as described in the literature [4, 23]. The patients



Legend: PSG: Polysomnography; ESS: Epworth Sleepiness Scale; Snoring Scale: Visual Analog Scale to Subjective Evaluation of Snoring; Quest. CPAP: Clinical Questionnaire of CPAP Clinical of the Sleep Institute in the Universidade Federal de São Paulo; MFTH: Myofunctional Therapy Assessment; CONSENT.: Consent and Signed a Waiver; Ex Diary.: Exercises Diary; Combination: CPAP+Myofunctional Therapy; PSG -CPAP: Polysomnography to adjust CPAP pressure.

Fig. 1 The experimental design

were instructed by one single speech pathologist to perform the following tasks:

Soft palate: elevation of the soft palate and uvula while pronounce an oral vowel intermittently /a/ (isotonic exercise) and continuously (isometric exercise). The palatopharyngeal, palatoglossus, uvula, tensor veli palatini, and levator veli palatini muscle are recruited in this exercise. The isotonic exercise also recruits pharyngeus lateral wall. These exercises had to be repeated daily, three times a day, for 3 min and were performed once a week under supervision to ensure adequate effort.

Tongue: (1) Brushing the superior and lateral surfaces of the tongue while the tongue is positioned in the floor of the mouth (5 times each movement, three times a day); (2) push the tip of the tongue against the hard palate and slide the tongue backward (20 times, three times a day); (3) suck the tongue upward against the palate, pressing the entire tongue against the palate (20 times); and (4) tongue rotation in the oral vestibule 10 times right and left side (5) forcing the back of the tongue against the floor of the mouth while keeping the tip of the tongue in contact with the inferior incisive teeth (20 times, three times a day).

Facial: (1) recruitment of the buccinator muscle against the finger that is introduced in the oral cavity, pressing the buccinators muscle outward (10 times each

side, three times a day). (2) Suck air from a syringe of 20 ml (five times, three times a day).

Stomatognathic functions:

1. Suction: (1) suck the yogurt with a narrow straw.
2. Breathing and speech: (1) forced nasal inspiration and oral expiration in conjunction with phonation of open vowels, while sitting; and (2) balloon inflation with prolonged nasal inspiration and then forced blowing, repeated five times without taking the balloon out of the mouth.
3. Swallowing and chewing: alternate bilateral chewing and deglutition, using the tongue in the palate, closed teeth, without perioral contraction, whenever feeding. This exercise aims for the correct position of the tongue while eating and targets the appropriate functionality and movement of the tongue and jaw. The patients were instructed to incorporate this mastication pattern whenever they were eating.

Both therapies were performed at home for 3 months with three daily exercise sessions of 20 min each. An exercise diary was completed. The patients were contacted weekly by the same examiner who evaluated whether the exercises were adequately performed and collected the diary containing

information about the performance of the exercises. The patients from the myofunctional therapy and combined groups received a new series of myofunctional therapy exercises and filled out a new exercise diary every week.

Adherence to placebo and myofunctional therapy interventions was assessed based on the percentage of time that the patients performed the exercises during the 3 months of treatment.

Continuous positive airway pressure treatment

All patients from the CPAP and combined groups underwent a full night, attended PSG to manually determine the optimum pressure of CPAP. The CPAP titration was performed according to the protocol proposed by the American Academy of Sleep Medicine [24], in order to abolish all abnormal respiratory events during sleep (apneas, hypopneas, arousals by increased respiratory effort, limitations on airflow and snoring). One CPAP device (REMstar® Plus; Respironics Inc., Murrysville, PA) with nasal mask, without humidifier, which was set to the optimal pressure according to the PSG, was provided to each patient. A coupled pressure hour meter was used to objectively evaluate the time of CPAP use in hours per day. The patients were monitored by the educational program with regard to CPAP use and were given guidelines on the disease in three visits (first week, first month, and third month).

Adherence to CPAP was defined by average number of hours of daily CPAP use. Patients who used the device for at least 4 h per night on 70% of the nights were considered to have good adherence.

Statistical analysis

The statistical analysis was performed using SPSS 18.0 software and Statistic software (version 7.0; StatSoft, Inc., Tulsa, OK, USA). The Kolmogorov-Smirnov normality test was used. Data are presented as means and standard deviations. For variables that were not normally distributed, the results were standardized using the Z-score.

To compare the groups, the general linear model (GLM) of repeated measures test was used, and when appropriate, the a posteriori Tukey's honest significant difference (HSD) test was used. The results of these comparisons were controlled for the effect of the basal AHI.

The paired samples *t* test (Student's *t* test) was performed. The Spearman correlation coefficient was used to correlate delta values between measurements.

The G*Power 3.1.10 program was used to calculate sample sizes. With an expected difference in the hours of CPAP used between the CPAP group and the combined group (0.40), α of 0.05 and sample size of 54 patients (27 patients in CPAP and

27 combined group), a power of 0.85 was found. The significance level was set at 5%.

Results

The flowchart of patient selection during this study started with 140 patients with 40 patients failing to complete the study. The 100 patients who finished the study protocol had been distributed to placebo group ($N = 24$), myofunctional therapy group ($N = 27$), CPAP group ($N = 27$), and combined group ($N = 22$). The final sample consisted of 100 men aged 48.1 ± 11.2 years old (mean \pm standard deviation), with a BMI of 27.4 ± 4.9 kg/m², an ESS score of 12.7 ± 3.0 , and an AHI of 30.9 ± 20.6 .

The data from the patients who did not complete the study were similar to the data from the patients in the final sample, except for age, which was lower in the group that did not complete the study (Table 1).

Table 2 shows that significant differences regarding age, BMI, and neck circumference were not observed between the groups. Compared with before treatment, the myofunctional therapy, CPAP, and combined groups showed improved ESS scores after treatment ($p < 0.001$ each) and when compared with the placebo group ($p = 0.04$, $p < 0.01$, and $p < 0.01$, respectively). After the washout period, there was no change in subjective sleepiness in the CPAP and combined groups, but the ESS score decreased in the myofunctional therapy group. The assessment of the analog

Table 1 Characteristics of patients who completed the study compared with those of the individuals who did not complete the study

	Patients completed the study ($n = 100$)	Patients did not completed the study ($n = 40$)	<i>P</i> value
Age (years)	48.1 \pm 11.2	39.4 \pm 10.1	<0.001
BMI (kg/m ²)	27.4 \pm 4.9	28.1 \pm 3.0	0.40
ESS	12.7 \pm 3.0	12.1 \pm 1.8	0.13
AHI	30.9 \pm 20.6	30.4 \pm 20.2	0.89
SE (%)	84.1 \pm 9.9	81.8 \pm 13.1	0.24
S1 (%)	5.4 \pm 3.2	5.9 \pm 5.6	0.44
S2 (%)	57.9 \pm 9.7	56.6 \pm 12.9	0.49
S3 + S4 (%)	16.7 \pm 7.7	18.8 \pm 9.5	0.08
REM (%)	19.9 \pm 5.6	19.3 \pm 8.1	0.56
AI	27.4 \pm 15.8	25.0 \pm 15.8	0.51
SpO ₂ min (%)	81.9 \pm 7.8	83.5 \pm 6.8	0.27

Student's *t* test. The data are presented as the mean \pm standard deviation. Significant values are shown in italics

BMI body mass index, ESS Epworth sleepiness scale, AHI apnea-hypopnea index per sleep hour, SE sleep efficiency, S1 stage 1, S2 stage 2, S3 + S4 stages 3 plus 4, REM rapid eye movements, AI arousal index per sleep hour, SpO₂ min minimal saturation of oxyhemoglobin

Table 2 Demographic data and Epworth sleepiness scale, and snoring frequency and intensity for the placebo, myofunctional therapy, CPAP, and combination groups before treatment, after treatment, and after washout

Parameters	Placebo group (N = 24)			Myofunctional therapy group (N = 27)			CPAP group (N = 27)			Combination group (N = 22)			P value
	Before	After	Washout	Before	After	Washout	Before	After	Washout	Before	After	Washout	
Age (years)	42.9 ± 10.5	43.4 ± 10.4	43.4 ± 10.4	45.2 ± 13.0	45.5 ± 13.0	45.6 ± 13.0	46.4 ± 9.1	46.8 ± 9.2	46.9 ± 9.1	47.5 ± 10.9	48.0 ± 10.8	48.1 ± 9.1	0.33
Circ. (cm)	41.9 ± 3.7	41.9 ± 3.6	42.9 ± 3.7	41.6 ± 3.7	41.5 ± 2.3	41.9 ± 2.5	41.9 ± 3.9	41.9 ± 3.7	41.5 ± 3.4	42.4 ± 2.8	41.8 ± 3.5	41.7 ± 3.5	0.78
BMI (kg/m ²)	28.6 ± 4.0	28.3 ± 3.9	29.0 ± 4.0	25.0 ± 7.4	26.7 ± 2.9	26.9 ± 2.9	28.7 ± 3.3	29.5 ± 3.2	27.4 ± 6.9	27.9 ± 2.4	28.3 ± 2.6	28.2 ± 2.8	0.27
ESS	12.8 ± 3.1	12.2 ± 5.2 ^{###}	10.5 ± 5.1	13.7 ± 3.2	7.5 ± 3.7*	10.4 ± 4.3 ^{***}	12.0 ± 2.1	7.2 ± 3.6*	8.8 ± 4.4 ^{**}	12.0 ± 2.6	7.3 ± 5.7*	9.5 ± 6.3	<0.001
Snoring Frequency	9.1 ± 1.8	7.1 ± 3.2 ^{###}	7.2 ± 3.1	8.5 ± 2.3	4.9 ± 3.2*	5.4 ± 3.3 ^{**}	8.8 ± 2.1	3.1 ± 4.1*	8.3 ± 3.0 ^{***}	8.9 ± 4.1	3.9 ± 4.2*	6.8 ± 3.1 ^{***}	<0.001
Snoring Intensity	8.6 ± 2.2	6.2 ± 3.6 ^{###}	6.0 ± 3.1	7.7 ± 2.3	4.3 ± 2.8*	4.8 ± 3.1 ^{**}	8.1 ± 2.4	2.6 ± 3.6*	6.6 ± 3.1 ^{***}	8.5 ± 2.3	3.1 ± 3.2*	5.9 ± 2.7 ^{***}	<0.001

The data are expressed as the mean ± standard deviation. Repeated measures ANOVA with Tukey's *posteriori*. The results were controlled for the effect of the apnea-hypopnea index. Significant values are shown in italics

Circ. neck circumference, *BMI* body mass index, *ESS* Epworth sleepiness scale

*Before vs. after $p < 0.001$; **before vs. washout $p = 0.02$; ***after vs. washout $p < 0.01$; # placebo group (after) vs. myofunctional therapy group (after) $p = 0.04$; ## placebo group (after) vs. CPAP group (after) $p < 0.01$; ### placebo group (after) vs. combination group (after) $p < 0.01$

scale of snoring intensity and frequency showed significant improvement in the myofunctional therapy, CPAP, and combined groups after treatment ($p < 0.001$ each) and in the CPAP and combined groups when compared with the placebo group ($p < 0.01$ each). The myofunctional therapy group maintained the improvement in the snoring intensity and frequency after the washout period ($p = 0.02$), whereas these values returned to levels similar to pretreatment levels in the CPAP and combined groups.

The average adherence to treatment was 55% in the placebo group, 63% in the myofunctional therapy group, 30% in the CPAP group, and 65% in the combined group. Table 3 shows the average hours of CPAP use. Patients in the combined group spent more time using the device than patients in the CPAP group after 1 week ($p = 0.01$) and after 3 months ($p = 0.02$) of treatment. Furthermore, adherence to the CPAP device based on the number of days that the patient used the device for more than 4 h per night was evaluated; 30% of the patients from the CPAP group and 50% of patients from the combined group showed good adherence.

In both groups, the pre- and post-treatment CPAP pressure values did not show significant changes in therapeutic pressure after treatment (Table 4).

The objective assessment of sleep by PSG (Table 5) showed that the groups were similar in pre-treatment. After treatment, there was an improvement in AHI, minimum SpO₂, and arousal index (AI) in the CPAP group ($p = 0.04$, $p = 0.02$, $p = 0.04$, respectively) and in the combined group ($p = 0.02$, $p = 0.02$, $p = 0.02$, respectively). When compared with the placebo group, the combined and CPAP groups showed a significant reduction in AHI and AI ($p = 0.04$, $p = 0.02$, respectively). A significant reduction in AHI was also observed in the myofunctional therapy group when compared with the placebo group ($p < 0.001$). After the treatment period, the AHI decreased by 50% in the myofunctional therapy group, 87% in the CPAP group, and 89% in the combined group (Fig. 2).

The analysis (Table 6) of the MMI delta (normalized) and the delta values (increase) of the tongue muscles and soft palate strength showed a significant negative correlation in the myofunctional therapy and combined groups.

Table 3 Hours of CPAP use for the CPAP and combined groups after 1 week, 1 month, and 3 months of use of the device

	Hours of CPAP Use		
	CPAP group	Combination group	P value
1 week	3.8 ± 2.1	5.3 ± 1.8	0.01
First month	4.2 ± 2.1	5.0 ± 2.1	0.24
Third month	3.6 ± 1.8	5.1 ± 2.3	0.02

Student's *t* test. The data are presented as the mean ± standard deviation

Table 4 The therapeutic pressures of the polysomnographic CPAP titrations performed pre- and post-treatment in the CPAP and combined groups

	Therapeutic pressure (cm H ₂ O)		
	CPAP group	Combination group	<i>P</i> value
First PSG with CPAP	9.8 ± 2.2	9.1 ± 2.2	0.25
Second PSG with CPAP	9.6 ± 2.0	8.9 ± 1.6	0.21

Student's *t* test. The data are presented as the mean ± standard deviation

Discussion

This study compared the CPAP adherence analyzing CPAP alone and in association with myofunctional therapy in patients with OSAS. The results showed that the myofunctional therapy alone or added to CPAP significantly decreased excessive sleepiness, snoring, and AHI. Despite the fact that the myofunctional therapy did not reduce the level of required CPAP, it significantly increased adherence to CPAP treatment and improved upper airway (UA) muscle strength.

We previously published with the same patient sample that myofunctional therapy alone as well as in association with

CPAP might be an alternative treatment for the improvement of the quality of life reports in patients with OSAS [5]. Other studies have shown that myofunctional therapy alone, using exercise protocols different than the present study, improves snoring and AHI in patients with OSAS [4]. In the study by Guimarães and coworkers [4], there was a moderate association between changes in neck circumference with changes in AHI, suggesting that exercise can induce UA remodeling, increasing its patency during sleep. A study by Dantas and coworkers [25] showed increased collagen type I in patients with OSAS when compared with a control group which showed higher levels of collagen type III. The authors suggested that the delayed response of contraction and relaxation of pharynx muscles during the transition from inspiration to expiration may lead to an increased collapse in the UA region. Exercises that target the oropharynx region increase the strength of the oropharynx muscles, thus repositioning the tongue under anteroposterior stress, and help to reduce the collapse of the UA muscles. Myofunctional exercises of local muscular endurance aim to tone the muscle groups in the oropharynx, optimizing muscle strength and mobility, and adjust the position of the soft tissues (soft palate, pharyngeal constrictor muscles, suprahyoid muscles, tip and

Table 5 Objective data on sleep using polysomnography for the placebo, myofunctional therapy, CPAP, and combination groups before treatment, after treatment, and after washout

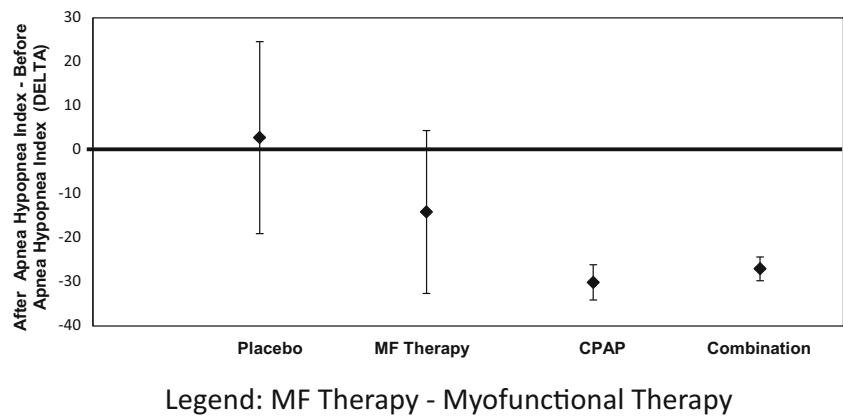
Parameters	Placebo group (<i>N</i> = 24)			Myofunctional therapy group (<i>N</i> = 27)			CPAP group (<i>N</i> = 27)			Combination group (<i>N</i> = 22)			<i>P</i> value
	Before	After	Washout	Before	After	Washout	Before	After	Washout	Before	After	Washout	
AHI	27.8 ±20.3	30.6***#.# ±21.8	27.8 ±15.0	28 ±22.7	13.9* ±18.5	21.3 ±21.4	34.4 ±22.4	4.3* ±4.0	29.7** ±25.4	30.4 ±19.8	3.4* ±2.7	29.6** ±25.1	<0.001
SE (%)	82.4 ±10.2	85.2 ±9.3	84.1 ±12.4	85.7 ±9.5	84.5 ±13.1	84.4 ±11.5	86.9 ±9.9	86.6 ±7.8	86.7 ±7.9	84.1 ±9.7	88.1 ±7.5	85.5 ±11.7	0.83
S1 (%)	5.5 ±3.7	5.5 ±4.6	4.8 ±3.1	4.8 ±3.8	5.1 ±3.7	4.5 ±3.8	5.2 ±3.1	5.7 ±3.6	6 ±3.8	4 ±2.3	4.1 ±2.0	4.5 ±3.2	0.8
S2 (%)	54.9 ±5.8	53 ±10.1	53.2 ±7.5	61.4 ±9.4	56.5 ±10.2	57.6 ±11.6	53.1 ±11.7	45.3 ±9.3	52.9 ±14.8	56.5 ±10.5	48.1 ±11.3	51.5 ±13.5	0.49
S3 + S4 (%)	16.5 ±5.4	19.9 ±11.7	19.8 ±8.5	14 ±7.5	18.5 ±11.0	16.9 ±8.5	18.8 ±9.0	25.2 ±7.4	18 ±10.6	18.7 ±8.2	25.4 ±7.8	22.2 ±11.0	0.08
REM (%)	21.1 ±5.8	21 ±5.6	20.2 ±6.8	17.9 ±5.4	18 ±5.9	19.2 ±7.0	20.9 ±5.6	22.4 ±6.5	21.5 ±7.2	19.1 ±5.8	20.6 ±7.1	19.7 ±6.2	0.86
AI	24.9 ±13.6	26.7#.# ±16.6	22.6 ±11.1	26.3 ±18.7	21.3 ±15.6	23.4 ±16.9	28.2 ±14.9	11.8* ±5.6	28.4** ±20.5	27.7 ±15.5	11.7* ±5.3	28.0** ±21.2	<0.001
SpO ₂ mean (%)	93.8 ±1.4	94 ±1.7	94 ±1.4	93.9 ±1.7	94.2 ±1.5	93.9 ±1.6	94 ±1.4	95.6 ±1.2	94 ±1.5	94.1 ±1.8	95.5 ±1.0	93.9 ±1.4	0.08
SpO ₂ min (%)	82.6 ±6.3	82.8 ±6.2	82.9 ±7.0	83.7 ±7.7	84.9 ±8.8	83 ±8.0	80.4 ±6.8	90.2* ±3.6	81.8** ±6.7	80.5 ±11.0	89.3* ±4.1	81.2** ±8.3	<0.001

The data are expressed as the mean ± standard deviation. Repeated measures ANOVA with Tukey's *posteriori*. The results were controlled for the effect of the apnea-hypopnea index. Significant values are shown in italics

AHI apnea-hypopnea index per sleep hour, SE sleep efficiency, S1 stage 1, S2 stage 2, S3 + S4 stages 3 and 4, REM rapid eye movement, AI arousal index per sleep hour, SpO₂ min minimal saturation of oxyhemoglobin, SpO₂ mean mean saturation of oxyhemoglobin

*Before vs. after *p* < 0.001; **after vs. washout *p* < 0.001; ***placebo group (after) vs. myofunctional therapy group (after) *p* < 0.001; # placebo group (after) vs. CPAP group (after) *p* = 0.04; ## placebo group (after) vs. combination group (after) *p* = 0.02

Fig. 2 Variation in the apnea-hypopnea index values after 3 months of treatment in the placebo, myofunctional therapy, CPAP, and combined groups. The data are presented as the mean \pm standard deviation



base of the tongue, cheeks, lips) and orofacial functions of chewing, sucking, swallowing, and breathing. These exercises may therefore reduce snoring and decrease the severity of OSAS [4].

Indirectly, an improvement in the MMI classification was observed in our study; this improvement was correlated with the increased strength of the tongue and soft palate during myofunctional evaluation, which was observed during wakefulness. This correlation could explain the improved objective sleep parameters that were observed during the night in patients with OSAS.

Our results suggest that there was an objective improvement in adherence to CPAP treatment as a benefit of myofunctional therapy. A limitation of this interpretation is that the myofunctional therapy and the combined groups were monitored more frequently than the CPAP group, which may have favored adherence. However, a recent study evaluated the influence of physical activity on adherence to CPAP use in patients with OSAS [26]. Ackel-D’Elia and coworkers [26] observed that during 2 months of treatment, there was no significant difference regarding adherence between the group that only used CPAP (4.7 h/night) and the group that used CPAP + exercises (4.2 h/night). In that study, the authors had the same limitation that the present study had. The

CPAP + exercises group was monitored three times a week, whereas the CPAP group was monitored only three times during the study; however, this difference did not influence adherence to the use of the device. Comparing the present study and the study by Ackel-D’Elia and coworkers [22], we assume that the effect of exercises that target the UA was the decisive factor for adherence to CPAP. Moreover, the patients were participating in an exercise program, which may have encouraged them to care more about their health and to be more attentive to the use of CPAP. This increased adherence in association with a series of specific exercises for the treatment of OSAS was observed in the present study during the first week of CPAP treatment. These data confirm previous findings suggesting that efforts to increase CPAP acceptance and use are performed mainly in the first 15 days of treatment because early acceptance and use determines the long-term use of this treatment [24].

The results of the present study showed that after myofunctional therapy with muscle exercises in the oropharyngeal region, there were no significant changes in CPAP therapeutic pressure when compared with the optimal CPAP pressure measured before treatment. This result could contradict the assumption that UA muscles would be strengthened by myofunctional therapy. Some authors associate the optimal

Table 6 Spearman correlation coefficient between the Modified Mallampati Index measures and tongue and soft palate muscle strength in the placebo, myofunctional therapy, CPAP, and combination groups

	Delta after-before treatment		Delta washout-before treatment	
	MMI vs. tension of tongue	MMI vs. tension of soft palate	MMI vs. tension of tongue	MMI vs. tension of soft palate
Placebo group	RHO -0.321 ($p = 0.13$)	RHO 0.094 ($p = 0.66$)	RHO 0.00 ($p = 0.99$)	RHO 0.36 ($p = 0.12$)
Myofunctional therapy Group	RHO -0.759 ($p < 0.001$)	RHO -0.716 ($p < 0.001$)	RHO -0.32 ($p = 0.10$)	RHO -0.17 ($p = 0.37$)
CPAP group	RHO -0.192 ($p = 0.34$)	RHO -0.302 ($p = 0.12$)	RHO 0.12 ($p = 0.59$)	RHO -0.002 ($p = 0.99$)
Combination group	RHO -0.998 ($p < 0.001$)	RHO -0.987 ($p < 0.001$)	RHO -0.24 ($p = 0.27$)	RHO -0.36 ($p = 0.09$)

MMI Modified Mallampati Index, RHO correlation coefficient r

CPAP pressure with the craniofacial aspects of patients with OSAS [27–29], and few studies have investigated the UA muscle response to CPAP. These results suggest that CPAP therapeutic pressure therapy depends on several complex and multifactorial conditions, and the myofunctional therapy approach addresses only a portion of these factors. The present study demonstrated that myofunctional therapy, alone or combined with CPAP, and CPAP treatment are all significantly effective in improving subjective sleepiness during the first month of treatment. Until now, the literature did not show any association between AHI and sleepiness [29]. However, the degree of excessive sleepiness is one of the components that determine OSAS severity [8, 14]. According to Kushida and coworkers [24], CPAP is indicated to improve subjective sleepiness in patients with OSAS. The present study showed that myofunctional therapy significantly reduced AHI and sleepiness and can be considered as an alternative OSAS treatment.

The subjective evaluation of snoring intensity and frequency showed significant improvement in the myofunctional therapy, CPAP, and combined groups when compared with the placebo group. The findings of the present study corroborate the previously described evidence. Complete abolition of snoring due to CPAP use is widely documented [24], and the subjective snoring improvement resulting from the use of exercises that increase UA muscle tone has also been described [4, 30–32]. However, after approximately 3 weeks without any intervention, the myofunctional therapy was more effective in maintaining the subjective improvement in snoring, although the reduced snoring frequency and intensity were greater in the CPAP and combined groups. The CPAP group would be expected to show no improvement in snoring after treatment withdrawal because this group did not receive specific muscle training.

The present study also evaluated the effect of myofunctional therapy and CPAP treatment on objective sleep parameters. As expected, both treatments, alone or combined, significantly decreased AHI when compared with the placebo group. These results support previous reports that showed positive AHI effects in response to CPAP treatment [26] and myofunctional therapy [4] when evaluated separately. The results did not, however, show an additional improvement in objective sleep patterns in patients with OSAS when myofunctional therapy was associated with conventional treatment, such as CPAP.

Myofunctional therapy is a therapeutic option for reducing the severity of apnea through exercises to tone the muscles of the oropharynx [4, 23], especially in mild to moderate cases. In severe cases, myofunctional therapy may give educational support and appropriate guidance as to the side effects during treatment with positive airway pressure devices [6, 7].

Our study has some limitations: (1) lack of objective assessment of orofacial muscles, as a magnetic resonance

imaging to evaluate the impact of the myofunctional exercises program on anatomy of UA; (2) limited duration of myofunctional therapy, since a longer time could show greater benefits in the objective sleep parameters; and (3) the exclusion of women in the group, since this population may also benefit from myofunctional therapy.

In summary, our study showed that after 3 months of training with a muscle exercise program that targeted the oropharynx of patients with OSAS, the sleepiness, frequency, and intensity of snoring and the apnea severity were decreased. Furthermore, when performed in association with CPAP, there was a significant increase in adherence to the use of the CPAP device during the first week of treatment. Thus, myofunctional therapy in OSAS patients may be considered as an alternative treatment and as an adjuvant intervention strategy for adherence to CPAP.

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Compliance with ethical standards The study was approved by the research ethics committee of the Universidade Federal de São Paulo—UNIFESP (CEP 2002/08) and registered at [ClinicalTrials.gov](https://www.clinicaltrials.gov) (NCT01289405). All patients gave written informed consent.

Conflict of interest The authors declare that they have no conflict of interest.

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References

- Schwab RJ, Gefter WB, Hoffman EA, Gupta KB, Pack AI (1993) Dynamic upper airway imaging during awake respiration in normal subjects and patients with sleep disordered breathing. *Am Rev Respir Dis* 148(5):1385–1400
- Mezzanotte WS, Tangel DJ, White DP (1992) Waking genioglossal electromyogram in sleep apnea patients versus normal controls (a neuromuscular compensatory mechanism). *J Clin Invest* 89:1571–1579

3. Weaver TE, Sawyer AM (2010) Adherence to continuous positive airway pressure treatment for obstructive sleep apnoea: implications for future interventions. *Indian J Med Res* 131:245–258
4. Guimarães KC, Drager LF, Genta PR, Marcondes BF, Lorenzi-Filho G (2009) Effects of oropharyngeal exercises on patients with moderate obstructive sleep apnea syndrome. *Am J Respir Crit Care Med* 179(10):962–966
5. Diaferia G, Badke L, Santos-Silva R, Bommarito S, Tufik S, Bittencourt L (2013) 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* 14(7):628–635
6. Smith I, Nadig V, Lasserson TJ (2009) Educational, supportive and behavioural interventions to improve usage of continuous positive airway pressure machines for adults with obstructive sleep apnoea. (2):CD007736
7. Weaver T, Grunstein RR (2008) Adherence to continuous positive airway pressure therapy: to challenge to effective treatment. *Proc Am Thorac Soc* 5(2):173–178
8. American Academy of Sleep Medicine (2005) International Classification of Sleep Disorders. 2nd edition: diagnostic and coding manual (ICSD-2). Westchester, IL
9. Johns MW (1991) A new method of measuring daytime sleepiness: the Epworth sleepiness scale. *Sleep* 14:540–545.
10. Allanson JE, O'Hara P, Farkas, LG, Nair, RC (1993) Anthropometric craniofacial pattern profiles in Down syndrome. *Am J Med Genet* 47:748–752.
11. Felício CM, Ferreira CLP (2008) Protocol of orofacial myofunctional evaluation with scores. *Int J Pediatr Otorhinolaryngol* 72(3):367–375
12. Rechtschaffen A, Kales A (1968) A manual of standardized terminology, techniques and scoring system for sleep stages of human subjects. Bethesda: National Institute of Neurological Diseases and Blindness. Neurological Information Network (Publication no. 204).
13. American Sleep Disorders Association (ASDA) (1992) EEG arousals: scoring rules and examples: a preliminary report from the Sleep Disorders Atlas Task Force of the American Sleep Disorders Association. *Sleep* 15(2):173–184
14. American Academy of Sleep Medicine (1999) Sleep-related breathing disorders in adults: recommendations for syndrome definition and measurement techniques in clinical research. The report of an American Academy of Sleep Medicine Task Force. *Sleep* 22(5):667–689
15. Cunali PA, Almeida FR, Santos CD, Valdrichi NY, Nascimento LS, Dal-Fabbro C, Tufik S, Bittencourt LRA (2011) Mandibular exercises improve mandibular advancement device therapy for obstructive sleep apnea. *Sleep Breath* 15:717–727
16. Fujii M, Logemann J (1996) Effect of a tongue-holding maneuver on posterior pharyngeal wall movement during deglutition. *Am J Speech Lang Pathol* 5:23–30
17. Logemann JA, Pauloski BR, Rademaker AW, Colangelo LA (1997) Speech and swallowing rehabilitation for head and neck cancer patients. *Oncology* 11(5):651–664
18. Furkin AM, Santini CS (1999) *Disfagias orofaríngeas. [Oropharyngeal dysphagia]*. São Paulo: Pró Fono.
19. Ferraz MCA (2001) *Manual Prático de Motricidade Oral: Avaliação e tratamento. [Practical Manual of Oral Motor Function: Evaluation and treatment]*. Rio de Janeiro: Revinter.
20. Krakauer LH, Di Francesco RC, Marchesan IQ (2003) *Respiração oral: abordagem interdisciplinar [Mouth breathing: an interdisciplinary approach]*. São José dos Campos, Pulso
21. Carvalho GD (2003) S.O.S. Respirador bucal—uma visão funcional e clínica da amamentação. [S.O.S. Mouth breather—a functional and clinical view of breastfeeding.]. Editora Lovise, São Paulo
22. Burger RCP, Caixeta EC, Dininno CQMS (2012) A relação entre apnéia do sono, ronco e respiração oral. [Relationship between sleep apnea, snoring and oral respiration.]. *Rev.CEFAC [online journal]* <http://www.cefac.com.br>
23. Ieto V, Kayamori F, Montes MI, Hirata RP, Gregório MG, Alencar AM, Drager LF, Genta PR, Lorenzi-Filho G (2015) Effects of oropharyngeal exercises on snoring: a randomized trial. *Chest* 148(3):683–691
24. Kushida CA, Littner MR, Hirshkowitz M, Morgenthaler TI, Alessi CA, Bailey D, Boehlecke B, Brown TM, Coleman J Jr, Friedman L, Kapen S, Kapur VK, Kramer M, Lee-Chiong T, Owens J, Pancer JP, Swick TJ, Wise MS, American Academy of Sleep Medicine (2006) Practice parameters for the use of continuous and bilevel positive airway pressure devices to treat adult patients with sleep-related breathing disorders. *Sleep* 29(3):375–380
25. Dantas DAS, Mauad T, Silva LFF, Lorenzi-Filho G, Formigoni GGS, Cahali MB (2012) The extracellular matrix of the lateral pharyngeal wall in obstructive sleep apnea. *Sleep* 35(4):483–490
26. Ackel-D'Elia C, da Silva AC, Santos Silva R, Truksinas E, Sousa BS, Tufik S, De Mello MT, Bittencourt LRA (2012) Effects of exercise training associated with continuous positive airway pressure treatment in patients with obstructive sleep apnea syndrome. *Sleep Breath* 16(3):723–735
27. Akashiba T, Kosaka N, Yamamoto H, Ito D, Saito O, Horie T (2001) Optimal continuous positive airway pressure in patients with obstructive sleep apnoea: role of craniofacial structure. *Respir Med* 95:393–397
28. Zonato AI, Bittencourt LRA, Martinho FL, Gregório LC, Tufik S (2006) Upper airway surgery: the effect on nasal continuous positive airway pressure titration on obstructive sleep apnea patients. *Eur Arch Otorhinolaryngol* 263(5):481–486
29. Nakata S, Noda A, Yagi H, Yanagi E, Mimura T, Okada T, Misawa H, Nakashima T (2005) Nasal resistance for determinant factor of nasal surgery in CPAP failure patients with obstructive sleep apnea syndrome. *Rhinology* 43(4):296–299
30. Puhan MA, Suarez A, Lo Cascio C, Zahn A, Heitz M, Braendli O (2006) Didgeridoo playing as alternative treatment for obstructive sleep apnoea syndrome: randomised controlled trial. *BMJ* 332(7536):266–270
31. Randerath WJ, Galetke W, Domanski U, Weitkunat R, Rühle K (2004) Tongue-muscle training by intraoral electrical neurostimulation in patients with obstructive sleep apnea. *Sleep* 27(2):254–259
32. Ojay A, Ernst E (2000) Can singing exercises reduce snoring? A pilot study. *Complement Ther Med* 8(3):151–156