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### **Authors**

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## REVIEW

# Myofunctional Therapy for the Treatment of Obstructive Sleep Apnea: A Narrative Review

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## Abstract

Obstructive sleep apnea (OSA) is a serious and prevalent public health disorder that can have long-term detrimental effects on the cardiovascular, nervous, and metabolic systems, and increase the risk of accidents and mortality. Newly identified pathophysiological traits of OSA include decreased upper airway pharyngeal dilator muscle responsiveness. Myofunctional therapy (MT) has been proposed as a treatment to improve dilator tone by performing isotonic and isometric exercises of the tongue and other upper airway muscles. This article reviews the beginnings of this therapy and describes systematic reviews and meta-analyses published until February 2023 focusing on our experience using MT as an isolated therapy for treating OSA in adult patients.

The search strategy was designed using the keywords obstructive sleep apnea, OSA; Sleep apnea; oropharyngeal exercises; MT; Tongue position; Meta-analysis; Systematic review; Assessment; Evaluation; Recommendations as referenced in PubMed, EMBASE, and the Cochrane Database of Systemic Reviews up to February 2023.

We then explain our personal views and publications on the matter, including our therapeutic applications of the Airway Gym. We describe the benefits of this procedure for patients and professionals in terms of the homogeneity of oral and oropharyngeal exercises; the ability to provide acoustic, visual, and mechanical biofeedback; the objective and easy control of the process; and the accuracy of the exercises performed.

**Keywords:** Myofunctional therapy, Narrative review, Obstructive sleep apnea, Oropharyngeal exercises, Updates on myofunctional therapy for sleep apnea

## 1. Introduction

According to the International Classification of Sleep Disorders, third edition, Obstructive Sleep Apnea (OSA) is characterized by repetitive episodes of apnea or hypopnea during sleep that leads to a decrease in blood oxygen saturation and usually end in short arousals from sleep [1]. Current literature estimates the prevalence of OSA as 4–30% of the world population [2]. This disorder has

serious consequences in terms of cardiovascular and neurovascular diseases [3,4], increased risk of traffic accidents [5], and other serious long-term outcomes, including mortality [6,7].

The management options for OSA include both conservative measures, such as continuous positive airway pressure (CPAP) [8], mandibular advancement devices (MAD) [9], weight reduction, avoidance of alcohol, sedatives, and sleeping in the supine position, and different surgical interventions

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[10]. OSA is a multifactorial disorder, and the current treatment options do not address many of the underlying causes.

CPAP is considered to be the gold standard treatment for almost every patient even though it is a palliative treatment when it is used and adherence is suboptimal [11,12].

Gou et al. published a meta-analysis in 2016 that included 18 studies of 4146 patients. They concluded that CPAP therapy was associated with a trend of decreased risk of cardiovascular events compared with a control group and with a nonsignificant decrease in the incidence of death and stroke. ESS and blood pressure were significantly lower in the CPAP group [13].

Traditionally, anatomical and craniofacial features are considered the single origin of this pathology. However, it is now known that other factors play an important role in the genesis of OSA. Four phenotypic causes have been described: anatomically low respiratory arousal threshold; oversensitive ventilatory control system; collapsibility of the dilating pharynx muscles during sleep, especially the genioglossus muscle, which moves posteriorly into the upper airway [14–16]. Dysfunction and imbalance of the oropharyngeal muscles play a significant role in maintaining pharyngeal airway patency [17,18].

Myofunctional therapy (MT) for OSA is considered an interesting tool for improving upper airway dilator muscle tonicity. MT uses isotonic and isometric exercises for the neuromuscular reeducation of the oral and oropharyngeal muscles to correct muscle imbalance and tonicity. This provides better stability of the stomatognathic system by promoting nose breathing and tongue resting against the hard palate. MT is useful for also improving the functions of breathing, mastication, swallowing, and speech [19].

### 1.1. Search strategy

We included a historic investigation into the beginnings of MT. Afterward, we did not attempt to present a meta-analysis or systematic review but a narrative review of meta-analysis and published reviews using PubMed, EMBASE, and the Cochrane Database of Systemic Reviews up to February 2023. A summary of each was conducted with a critical perspective. The search strategy was designed using the keywords Obstructive sleep apnea; Oropharyngeal exercises; MT; Tongue Position; Metanalysis; Systematic review as referenced in PubMed, EMBASE, and the Cochrane Database of Systemic Reviews (Fig. 1).

We excluded systematic reviews without a meta-analysis, articles about the use of MT to treat

pediatric OSA, those that evaluated only snoring, and those that included treatments in addition to MT.

Finally, we present a review of our considerations of the management of obstructive sleep apnea syndrome (OSA) from etiological and physiological perspectives based on MT.

### 1.2. Historical review

This discipline was born in the early 1900s when Cryer [20] and others in the American Orthodontics Society realized the close relationship between form and function. Rogers, Straub, Truesdell, Garliner [21], and other researchers thought that muscular forces can cause modifications of bone structures. They noticed that mouth breathers usually have a low lip tone and carry the tongue downward and forward, which pushes the teeth forward and results in malocclusion and other dental problems. Benno Lischer, an orthodontic pioneer, is credited with coining the term MT in 1912 [22].

In 1918, Alfred Rogers spoke to the EEUU national meeting of orthodontists about the importance of muscle balance relative to tongue position against the teeth [21].

Daniel Garliner, President of the Institute for MT in Coral Gables, Florida, considered that teeth are like sheep, which are continually being herded by the sheepdogs, the muscles. He was an important leader in this field who has promoted courses and published articles and books since 1965 about how to recognize orofacial muscle imbalance and suggested specific exercises to retrain the muscles of the tongue and face to correct the swallowing pattern. He said that MT should precede, be concomitant with, or be instituted after dental treatment has been completed, depending on the specific problem, and to avoid the recurrence of the problem [23,24].

In 1999 and 2003, Guimaraes et al. reported on the effects of phonoaudiological work on patients with OSA [25,26]. Puhan in 2006 reported that playing an Australian woodwind instrument, the didgeridoo, improved OSA significantly and proposed it as an alternative treatment for OSA [27].

In 2009, Guimaraes et al. described a complete regimen of isotonic and isometric exercises derived from speech therapy for the soft palate, tongue, lip, buccinator, and jaw muscles. These authors published a randomized controlled trial involving 31 patients with moderate OSA classified into two groups: a control group of 15 was given sham therapy and an exercise treatment group of 16 patients was given daily oropharyngeal exercises for 3 months. The apnea–hypopnea index (AHI)

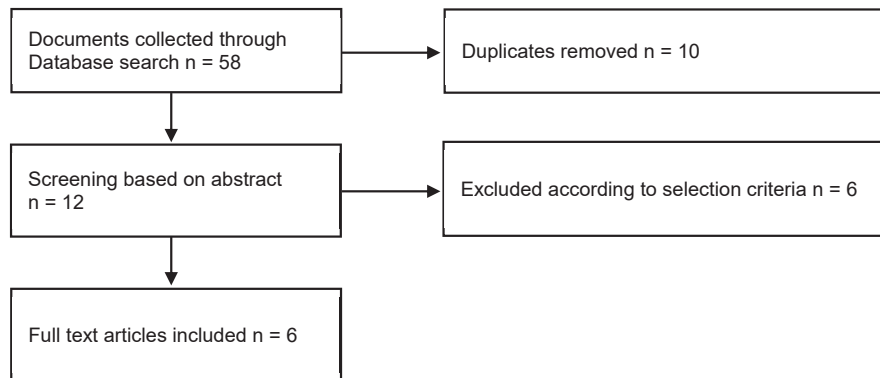


Fig. 1. Flow diagram demonstrating systematic reviews and meta-analysis of the efficacy of myofunctional therapy on obstructive sleep apnea in adults study selection. *n*, number of articles

decreased from  $22.4 \pm 4.8$  to  $13.7 \pm 8.5$  events/h in the exercise group. The exercise group also displayed a decrease in neck circumference, snoring, and daytime sleepiness, and improvement in the Pittsburgh Sleep Quality Index score [28].

### 1.3. Systematic reviews and meta-analysis of the efficacy of MT on OSA in adults

In 2015, Macario Camacho et al. presented a systematic review and meta-analysis that included nine adult studies with a total of 120 patients. They excluded studies that evaluated singing and instrument playing, observed a change in body weight of  $>10\%$ , assessed other interventions, or those that lacked objective data. The AHI decreased after MT from  $24.5 \pm 14.3$  to  $12.3 \pm 11.8$  events/h: mean difference = 14.26 [95% confidence interval (CI)  $-20.98$  to  $-7.54$ ;  $P < 0.0001$ ]. The AHI index decreased by about 50% in adults. The lowest oxygen saturation, intensity of snoring, and Epworth Sleepiness Scale (ESS) score improved. The heterogeneity of the oropharyngeal exercises and differences in the duration of daily execution and lack of long-term follow-up were mentioned as limitations [29].

Brien Hsu in 2020 conducted a systematic review and meta-analysis of studies that have evaluated MT, but this analysis included oropharyngeal exercises as well as speech therapy, breathing exercises, and playing the musical wind instrument, the didgeridoo. Nine randomized controlled trials with a total of 394 adults and children diagnosed with mild to severe OSA were included. All studies were considered to have a high risk of bias. The overall improvement in the AHI was 39.5%, and the author concluded that a statistically significant reduction in AHI can be achieved with MT. The limitations of this meta-analysis were that only nine studies were

included, the treatment duration ranged from 5 weeks to 4 months, and various types of exercises were evaluated, which resulted in some degree of heterogeneity [30].

Rueda et al. published a systematic review and meta-analysis of MT for treating OSA for the Cochrane Library in 2020 that included 347 participants in nine randomized clinical trials. They observed a large reduction in AHI (mean difference =  $-13.20$  points, 95% CI  $-18.48$  to  $-7.93$ ), reduced daytime sleepiness, and increased sleep quality. The authors concluded that although the results were promising, more trials involving blinding of participants, larger numbers of participants, and longer treatment and follow-up periods are needed [31].

In 2022, Meghpara et al. published a meta-analysis that included 15 studies of 237 patients in total given MT as an isolated treatment. The authors excluded studies in children and those that included singing, playing a musical instrument, or electrical stimulation. A reduction of 34% in AHI was found; the mean AHI decreased from  $27.99.0 \pm 16.16$  to  $18.56 \pm 13.1$  events/h. The mean difference was  $-10.47$  events/h, and the standard mean difference was  $-1.34$  (95% CI  $-0.84$  to  $-1.85$ ) ( $P < 0.00001$ ). Oxygen saturation improved by 2% and ESS score improved by 31% (a decrease of 4 points). This analysis also found a net reduction of 37% of the AHI in moderate OSA patients compared with 28% in severe OSA patients [32].

Zhang et al. published a meta-analysis of studies published until February 2022. Studies that used MT as a separate treatment for OSA in children (two studies, 25 patients) and adults (nine studies, 207 patients) were included. In the adults, the AHI scores decreased from  $26.8 \pm 10.0$  before to  $16.7 \pm 9.2$  events/h after MT. Eight adult studies (146 patients) reported that the lowest oxygen saturation increased from  $82.8\% \pm 4.8\%$  before to  $85.3\% \pm 55\%$  after MT.

The changes were significant in patients with mild and moderate OSA. Nonsignificant changes were observed in patients with severe OSA, which suggested that other therapies are also needed for this group [33].

Other systematic reviews and studies not included in a previous meta-analysis confirmed the promising results for MT for the treatment of OSA, for example, a 40–50% reduction in AHI [34–38].

#### 1.4. Personal experience of MT for OSA

Our investigations have noted that MT may be helpful for the management of patients with the hypotonic phenotype, who lack daily objective feedback from a speech therapist to ensure they are performing the exercises; these patients are likely to have poor compliance.

To address these problems, in 2017 we developed the first application (app) to treat OSA using MT. We initially named this app ‘apnea bye’ [39], but soon after changed the name to ‘Airway Gym’ [40].

This application comprises nine isotonic and isometric exercises that use proprioceptive training to strengthen oral and oropharyngeal muscles. Using the app, the patient interacts with the screen of a smartphone using a sensitive plug-in. In the app, each exercise is presented with a video, and each daily session takes about 20 min to complete.

This mobile health app provides acoustic, mechanical, and visual feedback about the accuracy of the exercises performed on a daily and monthly basis. Patients interact with their own smartphones at their convenience without having to attend to a speech therapist (Fig. 2).

We recommend doing the exercises 5 days a week for a minimum of 3 months to achieve results (Figs. 3 and 4). Patients can interact easily with their doctor via this app, through which they can send messages or ask for help. Another advantage is the low cost because the app does not require any other devices.

In 2019, we presented a case report of a patient with severe apnea who was nonadherent to CPAP. A floppy closing door epiglottis was confirmed during drug-induced sleep endoscopy. In this patient, the epiglottic collapse disappeared after 3 months of MT using the Airway Gym app, and the patient avoided surgery [40].

Eckert [14] described the different phenotypes of OSA and that poor upper airway muscle effectiveness is an important cause in many cases. The authors have aimed to find the ideal candidate for MT, that is, patients with the hypotonic phenotype.

In 2020, we reported on a prospective study involving 35 consecutive patients who were recently



Fig. 2. Practical example of management of Airway Gym app.

diagnosed with OSA and 20 controls [41]. We evaluated the genioglossus muscle tone, which was measured using the Iowa Oral Performance Instrument (IOPI) (Fig. 5), and somnography findings. The IOPI values were significantly lower in OSA patients ( $44.02 \pm 12.29$  and  $15.03 \pm 3.71$  kPa, respectively,  $P < 0.001$ ). IOPI tongue pressure and tongue collapse during drug-induced sleep endoscopy correlated significantly (Kruskal–Wallis  $\chi^2 = 25.82$ ;  $P < 0.001$ ).

Soon after, we reported the results of our new mobile health app Airway Gym used by a group of 20 patients diagnosed with apnea who were intolerant or nonadherent to CPAP or other treatments. Fifteen of the 20 patients completed the 90 sessions (75% adherence) and were compared with a control group of five patients who did not receive any

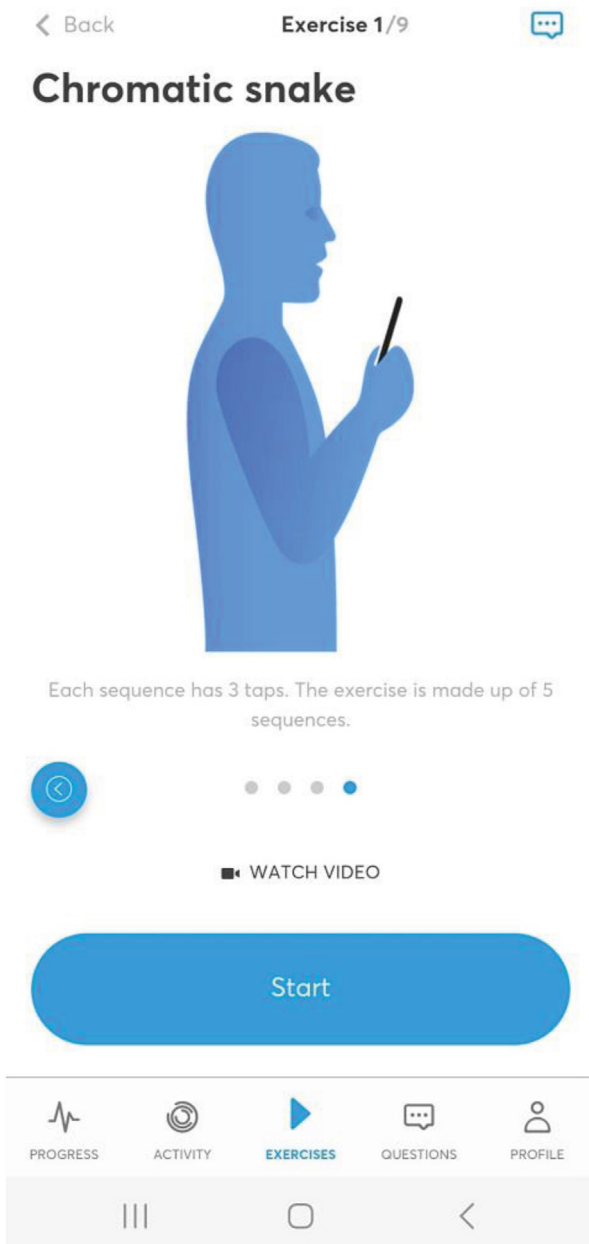


Fig. 3. Example of one of the exercises of Airway Gym app.

treatment. The AHI decreased in the therapeutic group from  $25.78 \pm 12.6$  to  $14.1 \pm 7.7$  events/h ( $P = 0.002$ ). There were no changes in the control group. Although the sample size was small, we consider that this app is a reasonable alternative for reluctant patients who do not respond to other therapies [42].

We conducted a prospective, randomized pilot trial to evaluate the effects of 3 months of MT with the Airway Gym app in patients with severe OSA. AHI decreased by 53%, from 44.7 (95% CI 33.8–55.6) to 20.88 (14.02–27.7) events/h ( $P < 0.001$ ). Maximum

peak tongue strength was tested, measured using the IOPI, increased from 39.83 (35.32–45.2) to 59.06 (54.74–64.00) kPa. AHI correlates significantly with the IOPI peak tongue strength (Pearson correlation coefficient  $-0.56$ ) [43].

A review of breathing reeducation and phenotypes of sleep apnea was published in 2021 in collaboration with Patrick Mckeown, a Buteyko Breathing Educator. This review examined each of the four phenotypes of OSA and explored how these could be targeted using breathing reeducation according to the three dimensions of functional breathing: biomechanical, biochemical, and resonant frequency [44].

Although the IOPI is an effective tool for measuring muscle strength for monitoring the efficacy of MT, it is too expensive for many physicians. In 2021, we reported on the use of the Tongue Digital Spoon (TDS) (Fig. 6) as an affordable tool for measuring genioglossus muscle strength [45]. When using the TDS, the patient presses the tip of the tongue against the spoon and presses the hold button on the handle when maximum effort is reached, which visualizes the data on the screen. The highest of the three measurements is considered the peak pressure. Twenty participants were evaluated using the TDS and IOPI. We found a significant correlation between these two tools (Pearson's correlation coefficient  $r = 0.69$ ,  $P < 0.001$ ). We concluded that the TDS is an economic and validated tool for monitoring tongue strength and helps identify hypotonic patients and monitor the results of MT.

We published a research protocol for a case–control clinical trial in 2021 to distinguish the muscular features in patients with severe apnea versus healthy controls with a view to optimize the future selection of patients for MT [46].

To assess the adherence to and efficacy of the Airway Gym app, we enrolled 65 patients who refused to use CPAP and any other treatment. Telemedicine allowed us to monitor the patients' use of this app objectively, and the adherence rate was 64.8%. After 90 sessions, AHI improved significantly from  $32.97 \pm 1.8$  to  $21.9 \pm 14.5$  events/h ( $t = 6.360$ ,  $P < 0.05$ ). Tongue peak pressure measured using the IOPI increased from  $44.4 \pm 11.08$  to  $50.66 \pm 10.2$  kPa ( $t = -3.8$ ,  $P < 0.05$ ) and correlated significantly with the AHI scores (Pearson  $r = 0.4$ ,  $P = 0.01$ ). Tongue mobility measured using the Hazelbaker score correlated significantly with the IOPI and AHI scores. We concluded that the Airway Gym is a useful tool and that it is essential to identify the presence of tongue-tie and to release it surgically before starting MT or other treatments [47].

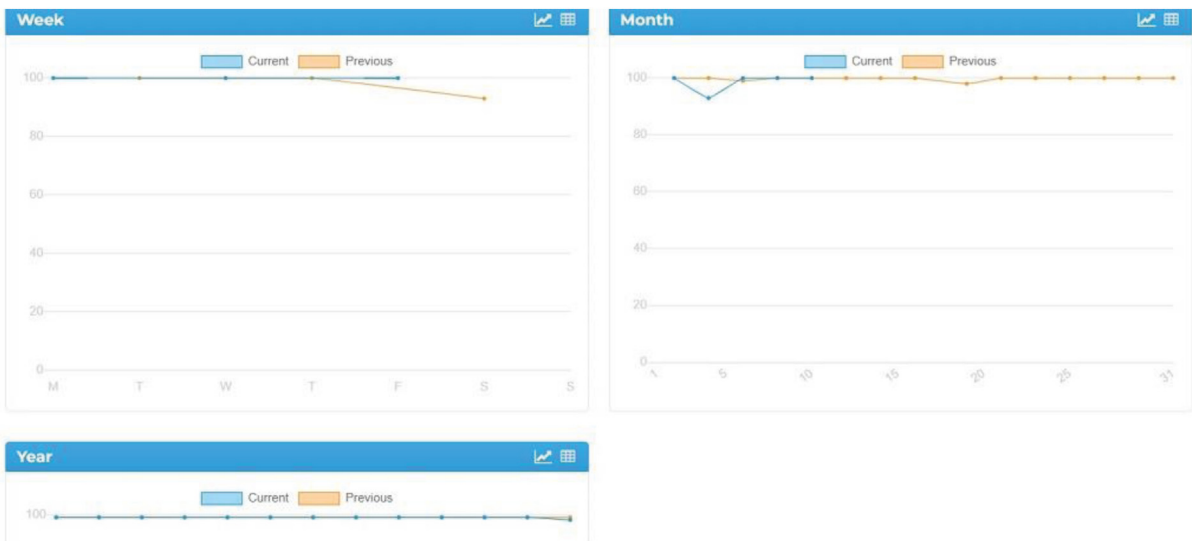


Fig. 4. Airway Gym provides information about progression in time of exercises performed.

Guilleminault et al. described lingual apraxia and impairment of stereognosis in OSA patients [48]. To evaluate this further, we conducted a prospective, nonrandomized pilot study of 25 patients with moderate to severe sOSA and 20 healthy controls. We assessed the IOPI scores and results of tests for lingual apraxia and stereognosis. Only 30% of patients with OSA complied with the apraxia test versus 100% in the healthy group. In the stereognosis test, the mean number of figures recognized was  $2.6 \pm 2.2$  in the OSA group versus  $5.7 \pm 0.9$  in the control group. Stereognosis test and IOPI score improved after 3 months of MT using the Airway

Gym app. We concluded that OSA patients have a deficit in sensorimotor function [49].

In 2021, we conducted a state-of-the-art review of the literature on the effectiveness of MT for treating sleep-disordered breathing. We examined the available evidence on the positive effects of reducing the severity of OSA as reflected in changes in polysomnographic parameters and clinical manifestations in adults [50].

In 2022, we used the TDS and IOPI to monitor the evolution of tongue strength in OSA patients before and after MT. We found significant improvements using both instruments and a high correlation between them [51].



Fig. 5. Iowa Oral Performance Instrument (IOPI) for tongue strength measure.



Fig. 6. Tongue digital spoon (TDS).



Despite the consensus about the essential contribution of hypotonia of the dilator upper airway muscles during sleep, a recent review of current official guidelines for treating apnea showed that only two guidelines consider MT as a treatment [52]. In 2021, the European Respiratory Society published a guideline for non-CPAP therapies that considered MT as an alternative treatment for patients reluctant to use CPAP or undergo surgery [53]. The latest Spanish Society of Pneumology and Thoracic Surgery guidelines published in 2022 suggest offering MT in three contexts: as an alternative treatment for mild and moderate OSA, as an adjunctive to CPAP to improve its efficacy and tolerance, and as complementary to MAD [54]. Only the American Academy of Dental Sleep Medicine recommends the evaluation of tongue mobility and the lingual frenulum. The latest algorithm of this society emphasizes that static causes of severe obstruction such as grade 4 tonsil size, important maxillary deformities, obstructive tongue base, etc need surgical intervention before any other treatment.

We have recently conducted a prospective case–control study to assess the weakness of the genioglossus muscle in patients with severe OSA. Multivariate regression revealed that an IOPI score <48 kPa was associated with severe OSA; the adjusted odds ratio (OR) was 9.96 (95% CI 2.5–39.1,  $P = 0.001$ ), specificity was 0.72, sensitivity was 0.79, and area under the curve (AUC) was 0.82. Severe OSA was also significantly associated with a TDS score <201 g/cm<sup>2</sup> (OR 27) (95% CI 4.74–153.6,  $P = 0.0001$ ); the specificity was 0.66, sensitivity was 0.93, and AUC was 0.86 [55].

We consider MT as an economical therapy with proven efficacy for reducing the AIH and daytime sleepiness and increasing oxygen levels without causing secondary effects that should be offered to patients with mild to moderate OSA. We agree with Wong et al. [56] that the hypotonic phenotype is a contraindication for upper airway surgery.

Isometric and isotonic exercises can change the pathophysiology of upper airway dilator muscles increasing their tone, confirmed by IOPI in kps [43] and TDS [51], and improving proprioceptive deficit with sensorimotor rehabilitation [49]. The nine exercises included in Airway Gym app, performed once a day for at least 20 min 5 times a week for 3 months have scientifically demonstrated its efficacy [42].

### 1.5. Future perspectives

More clinical trials are needed to guide decision-making for patients and professionals. These studies

should also include personalization of treatment and justification for the selection of specific exercises depending on the functional or anatomical origin of apnea in each patient. The studies should have an extended follow-up and include improved homogeneity of the exercises offered.

### Author name contributions

Maria Teresa Garcia-Iriarte: article writing, article screening, bias assessment; Carlos O'Connor-Reina data analysis, article drafting, and critical revision; Laura Rodriguez-Alcala: article screening, data analysis and collection, and manuscript drafting; Carlos O'Connor-Garcia: critical revision; Peter Baptista: article conception; Guillermo Plaza: article conception, article screening, bias assessment, manuscript drafting, and critical revision.

All authors have read and agreed to the published version of the manuscript.

### Conflicts of interest

COR is the designer of the application Airway Gym for MT. The rest of the authors have no conflict of interest.

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