Received: 15 November 2020 | Revised: 21 July 2021 | Accepted: 12 August 2021

DOI: 10.1111/joor.13247

ORIGINAL ARTICLE

JOURNAL OF ORAL REHABILITATION WILEY

Sensorimotor tongue evaluation and rehabilitation in patients with sleep-disordered breathing: a novel approach

Laura Rodríguez-Alcalá[1,2] | Juan Martín-Lagos Martínez[1] | Peter Baptista[3] | Raquel Ríos Fernández[4] | Francisco Javier Gómez[5] | Jesús Parejo Santaella[6] | Guillermo Plaza[7]

[1]Department of Otorhinolaryngology. Hospital, Universitario Clínico San Cecilio, Granada, Spain

[2]PhD Programme in Clinical Medicine and Public Health, University of Granada, Granada, Spain

[3]Department Otolaryngology, Clinica Universitaria de Navarra, Pamplona, Spain

[4]Autoinmune Disease Unit. Department of Medicine, Hospital Universitario San Cecilio, Granada, Spain

[5]Universidad de Granada, Granada, Spain

[6]Department of Radiology. Hospital, Universirario Virgen de las Nieves, Granada, Spain

[7]Department of Otorhinolaryngology. Hospital Sanitas La Zarzuela & Hospital Universitario Fuenlabrada, Universidad Rey Juan Carlos, Madrid, Spain

Correspondence
Laura Rodríguez-Alcalá MD. Department of Otorhinolaryngology. Hospital Universitario Clínico San Cecilio. Av. Conocimiento s/n 18016, Granada, Spain.
Email: laurarod3108@gmail.com

Abstract

Study objectives: To evaluate tone, apraxia and stereognosis dysfunctions in patients with SDB compared with healthy controls, and to monitor the effectiveness of Airway Gym® as an easy-to-use myofunctional therapy (MT) modality in terms of the tongue's motor and sensory responses, comparing results before and after therapy.

Methods: This was a prospective, non-randomised pilot study of 25 patients with moderate to severe obstructive sleep apnoea-hypopnoea syndrome (OSAHS), 25 patients with primary snoring (PS) and 20 healthy controls. Qualitative and quantitative instruments—Iowa Oral Performance Instrument (IOPI), lingual apraxia and stereognosis tests were used to assess tongue sensorimotor function.

Results: 22 patients with PS, 21 with OSAHS and all 20 controls ended the therapy. In OSAHS, the Epworth Sleepiness Scale score decreased from 16 $\pm$ 7.3 to 12 $\pm$ 4.5 after therapy ($p = 0.53$). In PS and OSAHS groups, the IOPI scores increased significantly. These measures did not change significantly in the controls. Lingual apraxia testing showed that controls performed all the manoeuvres, whereas PS 5.6 $\pm$ 1.4 and OSAHS 4.5 $\pm$ 1.9 ($p = 0.14$). In the stereognosis test, the mean number of figures recognised was 2.6 $\pm$ 2.2 in OSAHS, 3.3$\pm$1.2 in PS and 5.7$\pm$0.9 in control group ($p < 0.05$). Patients with OSAHS recognised circles and ovals less often.

Conclusion: Using the Airway Gym®app produced improvements in sensorimotor tongue function in patients with SDB, due to continuous stimulation of the brain based on proprioceptive training required to localise responses when doing the exercises.

KEYWORDS
apraxia, exercise, myofunctional therapy, obstructive sleep apnoea-hypopnoea syndrome, oropharynx, sleep apnoea, stereognosis

1 | INTRODUCTION

Obstructive sleep apnoea-hypopnoea syndrome (OSAHS) is a very prevalent disease, affecting 1%–3% of children, 6%–20% of men and 6%–9% of women in middle age. Advanced age, male sex and the body mass index (BMI) are the most important risk factors.[1]

The causes of OSAHS are not fully understood. A multifactorial origin is suggested, where anatomical and functional factors interact, promoting the collapse of the upper airway, as a consequence of an imbalance between the forces that tend to close it and those that keep it open.[2,3] The pioneering studies of Remmers et al.[4] concluded that the malfunctioning of the upper airway's dilating muscles causes the obstruction and pathogenesis of the disease, with the main dilator muscle being the genioglossus. White[3] proposed that neuronal injury affects the muscles of the upper airways from repetitive exposure to intermittent hypoxia and/or mechanical stress as a result of snoring and recurrent closure of the upper airways that contribute to the progression of OSAHS. Guilleminault et al.[5] described lingual apraxia and impairment of stereognosis in patients with OSAHS, including children and adults. Lingual apraxia is the inability to perform certain movements. Lingual stereognosis is the inability to identify certain shapes with the tongue inside the mouth. In their retrospective study, lingual stereognosis was found in 11.3% of adults and 18% of children with OSAHS. These findings were consistent with impairments in velopharyngeal sensibility reported previously.[6] They suggested that myofunctional therapy (MT) might help improve airway function in adults and children with OSAHS.

MT is based on orofacial exercises. It is a treatment modality applied to subjects with orofacial myofunctional disorders, which can interfere with the development or functioning of the above-mentioned structures. It demonstrates benefits by promoting changes in the upper respiratory tract's dysfunctional muscles and has been used successfully to reduce the severity of OSAHS and associated symptoms in adults.[7] MT has also been suggested for helping to reduce the apnoea-hypopnoea index (AHI) associated with snoring,[8] in improving the quality of life,[9] and as an adjuvant intervention strategy to support adherence to continuous positive airway pressure (CPAP) therapy.[10] However, few high-quality randomised studies, especially over the long term, have been reported.[11]

Villa et al.[12] found significant differences in labial and lingual tone values measured by the Iowa Oral Performance Instrument (IOPI) between children with sleep-disordered breathing (SDB) who received 2 months of MT and those who did not. They also found improvements in the habit of oral breathing and the oxygen desaturation index. De Felício et al.[13] also found significant differences in labial and lingual tone measured by IOPI between children with OSAHS and those with SDB compared with healthy controls. They concluded that children with OSAHS have an impaired motor coordination pattern of the oropharyngeal muscles. In adults, O'Connor-Reina et al. concluded that a series of 30 patients with severe OSAHS had lower tongue and lip strength tone measured with IOPI than did healthy controls. [14]

Statement of Significance

Myofunctional therapy is a theoretically effective treatment in patients with moderate OSAHS and an adjunctive treatment to continuous positive airway pressure therapy, but more scientific evidence is needed to establish its efficacy. Here, we measured sensorimotor tongue function (apraxia, strength and stereognosis) before and after oropharyngeal exercises performed with an application based on proprioceptive rehabilitation called Airway Gym®. We measured responses after performing exercises with this application in healthy controls, simple snorers and patients with OSAHS using different qualitative and quantitative instruments. We found lingual hypotonia, apraxia and impaired stereognosis in the patients with OSAHS. Improvements in the measurements and the patient's quality of life were observed after carrying out the exercises.

Buterbaugh et al. described a diminished change in brainstem activity during swallowing and reduced cerebrovascular reactivity in patients with OSAHS.[15] This presumably translates into adverse brain sensorimotor consequences from potential grey substance loss and is likely associated with adverse cerebrovascular consequences. These sensorimotor deficits worsen the functions of upper airway muscles, promoting lower tone, impaired stereognosis and apraxia.[6]

Proprioceptive training based on tactile, acoustical and visual feedback can yield meaningful improvements in somatosensory motor dysfunction found in patients with cerebrovascular deficits.[16] A novel mHealth application ('app' for short), Airway Gym®, has been designed to perform MT to promote oropharyngeal exercises based on proprioceptive training while interacting with a smartphone. It provides a useful tool that allows the performance of a series of exercises using the smartphone. It facilitates feedback to the patient and the physician of compliance with the therapy. The proposed average training time is 15 min a day, 5 days per week. The Airway Gym® app includes exercises to strengthen the tone of the lingual and pharyngeal muscles, which are mainly responsible for the collapse of the upper airway during sleep.[17-19] Furthermore, several works have demonstrated the effectiveness of the Airway Gym®, improvement the sleep apnoea hypopnea index (AHI). [17-20]

The objective of the study was to evaluate tone, apraxia and stereognosis dysfunctions in patients with SDB compared with healthy controls, and to monitor the effectiveness of Airway Gym® as an easy-to-use MT modality in terms of the tongue's motor and sensory responses, comparing results before and after therapy.

2 | METHODS

A prospective non-randomised pilot study was designed, including 25 individuals with primary snoring (PS), 25 patients with moderate-to-severe OSAHS and 20 healthy control subjects. Our local ethics committee approved the study (AWGAPN-2019-01), and all participants gave written informed consent.

2.1 | Sample size calculation

The effectiveness of the use of the app for performing MT in patients with sensorimotor deficits was evaluated using the percentage changes in the IOPI observed during follow-up as the primary outcome measure. This percentage was calculated from results reported in previous studies of this app.[17,19] Based on an alpha level of 0.05 and power of 0.80, we estimated that 60 participants would be required. The sample size was calculated using XLSTAT (v. 16, Addinsoft France).

2.2 | Patients

Patients were tested using in-office examinations, sleep questionnaires, polysomnography (PSG), motor evaluation of the oropharyngeal muscles, lingual stereognosis evaluation and quantitative instruments measuring the muscle tone of the genioglossus and buccinator muscles with the IOPI tool, before and after 3-month therapy with the AirwayGym® app. Eligible patients were those aged 18–75 years who were evaluated in the Sleep Laboratory of the Pneumology Department of a university hospital.

2.2.1 | Inclusion and exclusion criteria

We included patients with a recent diagnosis of primary snoring (PS) (AHI<5) or moderate to severe OSAHS (AHI >15), full dentition and grade I–IV tonsils. We excluded patients with one or more of the following conditions: age <18 and >75 years, BMI of 32 kg/m^2 or greater, severe obstructive nasal disease, ankyloglossia, inability to complete questionnaires, severe alcoholism, unstable coronary heart disease, regular use of hypnotic medication, a history of stroke, craniofacial malformations, hypothyroidism, neuromuscular diseases, heart failure, coronary artery disease or systemic diseases associated with a known inflammatory state (eg arthritis, vasculitis or sarcoidosis). Furthermore, patients with a history of rehabilitative treatment of the orofacial musculature as well as any prior therapy for OSAHS (surgery or mandibular advancement device) that might have influenced the response to MT were also excluded.

2.2.2 | Otolaryngology examination

At the initial visit, participants were evaluated by an ear, nose and throat (ENT) specialist and underwent rhino-oro-fibrolaryngoscopy, Friedman staging, Marchesani protocol[21] and examination of their temporomandibular joint.[22] In addition, anthropometric variables were measured (weight, height, BMI, neck and waist circumference) pre- and post-intervention.

Tongue manoeuvres at the initial examination

1. Pull your tongue straight out of the mouth as far as you can
2. With your tongue out, use it to touch the right cheek
3. With your tongue out, use it to touch the left cheek
4. Place the tip of the tongue in the middle of the roof of your mouth
5. Place your tongue on the top of your upper teeth
6. With your tongue out, use it to try touching your nose

FIGURE 1 Tongue manoeuvres (six types) assessed at the initial examination

2.3 | Questionnaires

2.3.1 | Epworth sleepiness scale (ESS)

The ESS was used at the beginning and end of the study to assess any subjective changes in the disease's main symptoms.[23]

2.3.2 | Visual analog scale (VAS)

A VAS was used to quantify snoring changes as assessed by the sleeper's partner at the beginning and end of the study.

2.3.3 | Nasal obstruction and septoplasty effectiveness (NOSE) scale

This scale was used to assess any nasal symptoms before starting the study.[24] Patients with severe symptoms (NOSE >70%) were excluded, and optimisation surgery was proposed to them.

2.4 | Sensorimotor evaluations

2.4.1 | Apraxia testing

Patients were asked to perform specific manoeuvres with the tongue as outlined (Figure 1). These manoeuvres were performed twice. If the subject was unable to perform a manoeuvre after a visual demonstration, the test was scored as abnormal. These manoeuvres have been reported previously, as they are used in MT.[5]

2.4.2 | Figure testing using the Profono intraoral stereognosis instrument (ISI)

In the figure test, we showed the patient a series of shapes: circle, half-circle, oval, rectangle, triangle and square. The patient was later blindfolded and asked to identify each figure placed individually on the tongue for 10 s. The test was repeated up to three times if needed.[5]

2.4.3 | IOPI

This test was used to measure variables related to tongue function. A technical description of the IOPI is provided in the reference manual: 'The instrument is a pressure transducer connected to a battery-operated amplifier, signal conditioning circuit, and digital voltmeter'. A peak holding circuit displays peak pressure in kPa on a digital readout. We measured the variables up to three times and recorded the highest value.[25]

2.5 | Laboratory procedures

Standard laboratory polysomnography (Somté PSG, Compumedics Ltd., 2006, Abbotsford, Australia) was performed according to the technical specifications of the American Academy of Sleep Medicine.[26] The recorded variables were obtained using electroencephalography (C3-A2, C4-A1, O1-A2 and O2-A1), electrooculography (two channels), chin and leg electromyography and electrocardiography. Frontal electrodes were not used. Respiratory variables were measured using linearised nasal pressure prongs and the flow waveform of the oronasal thermal signals. Respiratory effort signals were measured through inductive bands that recorded ribcage and abdominal movements. Oxygen (O_2) saturation, body position and snoring were also recorded. Apnoea and hypopnoea were analysed and scored according to the following criteria: hypopnoea was defined as a ≥30% decrease in airflow signal amplitude lasting ≥10 s and accompanied by ≥3% O_2 desaturation. Apnoea was defined as a ≥90% decrease in airflow signal amplitude lasting ≥10 s. The oxygen desaturation index (ODI) was used to quantify O_2 desaturation ≥3%. The tests were used to define PS with an AHI<5 events/h of sleep, moderate OSAHS as an AHI of 15–29.9 events/h of sleep and severe OSAHS as ≥30 events/h of sleep.

Once the participants had been included in the study, AirwayGym® app therapy was started. The proposed average training time was 15 min/day, 5 days per week. It included exercises to strengthen the tone of the lingual and pharyngeal muscles, mainly responsible for the collapse of the upper airway during sleep.[17,18] Three months after using the app, a re-evaluation was done repeating the ENT in-office examination in order to evaluate any adverse reaction to exercises, sleep questionnaires, lingual stereognosis evaluation and IOPI measurements.

2.6 | Statistical analysis

The data were analysed using IBM SPSS Statistics Base 22.0 software (IBM Corp., Armonk, NY, USA). The Mann-Whitney non-parametric *U* test was used to analyse the variables praxis, gnosis and tongue tone among three groups of patients: OSAHS, PS and healthy. A value of $p < 0.05$ was considered statistically significant. The examiners evaluated the results prospectively and blinded to the participants' identities.

3 | RESULTS

In our final setting, we examined 21/25 (88%) patients with newly diagnosed moderate-to-severe OSAHS, 22/25 (84%) subjects with PS and 20 healthy controls after they met the inclusion and exclusion criteria of the study and had followed the exercises for 3 months. Four patients in the OSAHS group and three in the PS group did not end the study and discontinued the exercises.

3.1 | Anthropometric measures

Most of the patients included in our study were middle-aged men and slightly overweight, but none were considered to be obese. All 20 patients with OSAHS were using the CPAP device during the performance of the study. Baseline BMI, AHI and sleepiness assessment results are shown in Table 1. We did not find any significant change in the BMI, neck circumference or waist values after performing the exercises (Table 2).

3.2 | Lingual praxis manoeuvres

At the beginning of the study, patients were asked to perform lingual manoeuvres to assess tongue motor dysfunction, but we observed

TABLE 1 Characteristics of patients with primary snoring (PS), obstructive sleep apnoea (OSAHS) and healthy. Mean and standard deviation (SD)

	PS (N=22)		OSAHS (N=21)		*Healthy(N=25)*		
Variable	Mean	SD	Mean	SD	Mean	SD	p^a
Age (years)	49.7	7.9	51.4	9.3	44	6.4	0.57
BMI (Kg/m^2)	24.1	3.4	25.4	4.1	22,8	5.6	0.4
Neck (cm)	39.4	5.2	40.8	5.6	38.2	4.8	0.8
Waist (cm)	95.0	9.6	98.0	10.3	88	11.1	0.06
AHI (e/h)	1.1	0.27	21.8	6.3	<5	-	<0.001[*]

Abbreviation: BMI, body mass index.

[a]Non-parametric Kruskal-Wallis test. $p < 0.05$ difference statistically significant.

[*]represents the significant results in the statistical analysis for a p value <0.05.

TABLE 2 Variables analysed in the group of patients SDB before and after 3 months of treatment with the AirwayGym® app

	Pre-AirwayGym® Mean (SD) ($N = 22$)	Post-AirwayGym® Mean (SD) ($N = 22$)	p^a
Primary snoring (PS) group			
Epworth scale	9 (7.3)	7 (4.5)	>0.05
VAS scale	7 (2.5)	3 (1.2)	0.04*
ISI ($N = 6$ figures)	3.4 (1.2)	5.4 (0.6)	0.04*
Tongue manoeuvres ($N = 6$)	5.6 (1.4)	5.7 (0.9)	>0.05
IOPI tongue (kPa)	42 (16.7)	68 (12.4)	0.04*
IOPI lips (kPa)	29 (13.4)	45 (9.4)	0.03*
Neck (cm)	39.4 (5.2)	38.1 (7.3)	>0.05
Waist (cm)	95 (9.6)	92 (6.2)	>0.05
OSAHS group			
Epworth scale	15 (6.5)	11 (3.6)	>0.05
VAS scale	8 (2.5)	6 (1.2)	>0.05
ISI ($N = 6$ figures)	2.6 (1.2)	4.6 (1.2)	0.04*
Tongue manoeuvres ($N = 6$)	4.5 (1.9)	5 (0.8)	>0.05
IOPI tongue (kPa)	31 (19.5)	57 (14.2)	0.04*
IOPI lips (kPa)	21 (8.8)	37 (12.3)	0.05*
Neck (cm)	40.8 (5.6)	40.1 (7.3)	>0.05
Waist (cm)	98 (10.3)	95.6 (9.1)	>0.05

Abbreviations: IOPI, Iowa oral performance instrument; PS, primary snoring; SDB, Sleep-disordered breathing; VAS, scale, visual analogue scale.

ISI Figure testing using the Profono intraoral stereognosis instrument.

[a]Mann-Whitney test.

*represents the significant results in the statistical analysis for a p value <0.05.

different percentages of patients complying with this: all the controls complied, but only 65% of patients with PS and 30% of patients with OSAHS did so (Table 3). However, after performing the exercises with the Airway Gym® app, we found a modest improvement in the two groups of subjects with SDB, but this was not statistically significant; Table 2).

3.3 | Lingual stereognosis

Lingual stereognosis was significantly different between the three groups (Table 4). In the stereognosis test, the mean number of figures recognised was 2.6 ± 2.2 in the OSAHS group, 3.3 ± 1.2 in the snoring group and 5.7 ± 0.9 in the control group ($p < 0.05$). Patients with OSAHS recognised circles and ovals less often (Figure 2). However, after performing the exercises with the Airway Gym® app, we did not find significant improvement (Table 2, Figure 3).

3.4 | IOPI measurements

Tongue strength was statistically significantly different between the three groups (Table 5). The tongue's strength or muscle tone assessed with the IOPI tool improved significantly in both groups ($p = 0.034$) after treatment with the Airway Gym® app (Table 2, Figure 3). The values reached the reference values for the normal population listed in the IOPI manual. In the PS group, IOPI tongue scores changed from 42 ± 16.7 to 68 ± 12.4 kPa ($p = 0.04$), and the IOPI lip score changed from 29 ± 13.4 to 45 ± 9.4 kPa ($p = 0.03$). In the OSAHS group, the IOPI tongue score changed from 31 ± 19.5 to 57 ± 14.2 kPa ($p = 0.04$), and the IOPI lip score changed from 21 ± 8.8 to 37 ± 12.3 kPa ($p = 0.03$).

3.5 | Questionnaires

Three months after Airway Gym® app treatment (Table 2), we found significant improvement in VAS in the two groups with SDB (Figure 3). However, although the ESS improved too, it did not reach statistical significance. Thus, in the patients with OSAHS, the ESS score decreased from 16 ± 7.3 before to 12 ± 4.5 after therapy ($p = 0.53$).

4 | DISCUSSION

This pilot controlled non-randomised study is the first to analyse the lingual sensory and motor response in patients with SDB

Group	x̄	%	p^a
Control group (*N* = 20)	6.0 ± 0.0	100	Control vs PS group; >0.05
PS group (*N* = 21)	5.6 ± 1.4	93	Control vs OSAHS group; <0.05*
OSAHS group (*N* = 22)	4.5 ± 1.9	75	PS group vs OSAHS group; >0.05

PS, primary snoring; OSAHS, sleep apnoea-hypopnea syndrome.
[a]Mann-Whitney test.
*represents the significant results in the statistical analysis for a *p* value <0.05.

TABLE 3 Mean (x̄) and percentage (%) of the total number of tongue manoeuvres (*N* = 6) performed in the different study groups before treatment with AirwayGym® app exercises

	No. of figures	ISI test (*N* = 6)	p^a
Control group (*N* = 20)	5.7 ± 0.9		Control vs PS group; <0.045*
PS (*N* = 21)	3.4 ± 1.2		Control vs OSAHS group; <0.01*
OSAHS (*N* = 22)	2.6 ± 2.2		PS group vs OSAHS group; 0.06

Abbreviations: PS, primary snoring; OSAHS, sleep apnoea-hypopnea syndrome.
[a]Mann-Whitney test.
*represents the significant results in the statistical analysis for a *p* value <0.05.

TABLE 4 Evaluation of stereognosis with the Profono Intraoral Stereognosis Instrument (ISI) test in the healthy control, primary snoring (PS) and obstructive sleep apnoea-hypopnoea syndrome (OSAHS) groups before performing AirwayGym® lingual exercises

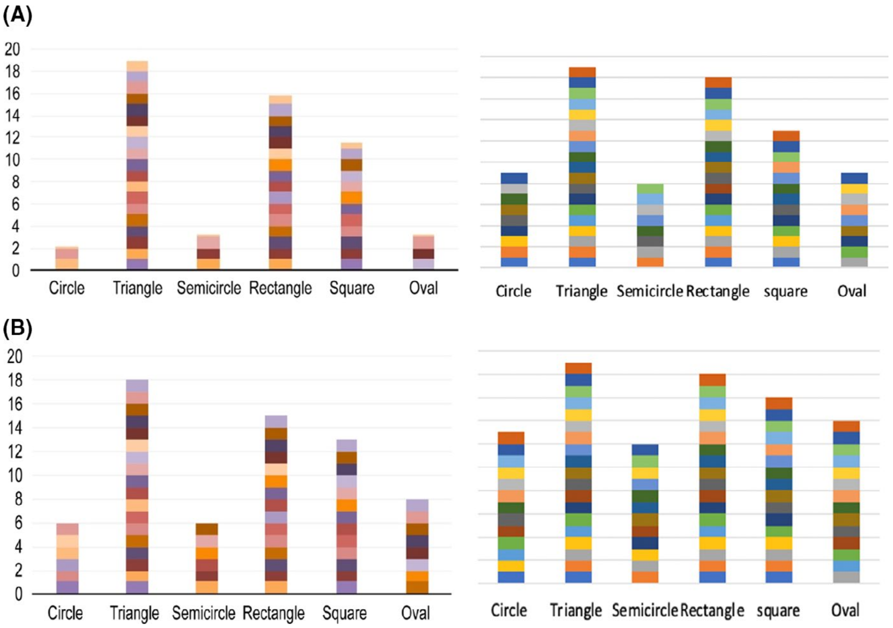

FIGURE 2 Representations of the mean shapes identified by patients with obstructive sleep apnoea-hypopnoea syndrome (OSAHS) or primary snoring (PS) using the Profono intraoral stereognosis instrument. (A) Shapes identified by the OSAHS group (*N* = 21) before performing the Airway Gym® exercises. (B) Shapes identified by the PS group (*N* = 22) before performing the Airway Gym® exercises.

before and after performing oropharyngeal exercises. We demonstrated an impairment in sensorimotor muscle function in these patients and its improvement after this therapy. We aimed to better understand the mechanisms involved in obstructive sleep disorder. Given our experience, upper airway muscles in patients with SDB should be evaluated from three aspects: praxis, stereognosis and tone.

As Eckert described, there is no doubt that a crowded, narrow or inherently collapsible upper airway is the key cause of OSAHS, but proper muscle activity is essential for speaking, swallowing and breathing.[27] Understanding the concepts of neural control, muscle responsiveness and muscle effectiveness helps explain how pharyngeal dilator musculature is involved in the pathogenesis of OSAHS.[27] The ability to respond to changes in CO_2 and pharyngeal pressure during sleep is lost in patients with OSAHS. More than one-third of such patients do not increase activity in the genioglossus muscle, which results in a poor protective response of the musculature to neural drive. One way to improve the upper airway's stability is to improve the coordination of the tongue with protrusion activities by improving the cortical excitability produced by the genioglossus muscle. [27]. We found that the ability to perform lingual manoeuvres was significantly worse in patients with SDB than that in healthy controls. However, after MT we could not find any significant improvements in this measure. We believe that this could be because we excluded patients with ankyloglossia or other anatomical limitations, and because of the small sample size.

We observed a significant improvement in the ISI stereognosis test response after patients completed the 3-month therapy with the Airway Gym® app. The advantage of MT using this app is that the patient's active training requires both mental and motor activation

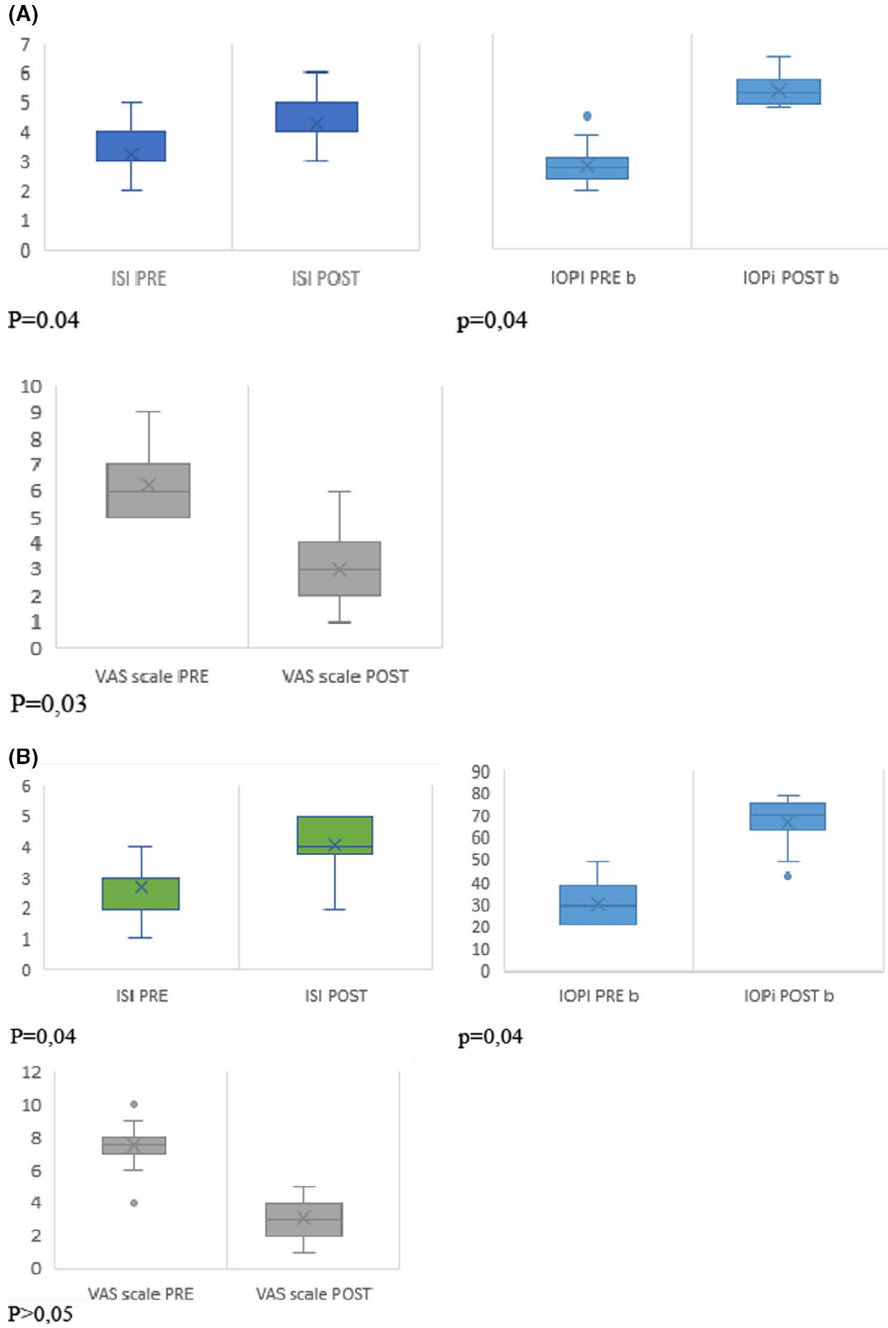

FIGURE 3 Box plot showing statistically significant differences before and after AirwayGym® exercises in the variables analysed, in the two groups of patients with DSB (*N* = 43). (A) Box plot showing statistically significant differences before and after AirwayGym® exercises in the variables analysed in the group of patients with primary snoring (PS) (*N* = 22). (B) Box plot showing statistical differences before and after AirwayGym® exercises in the variables analysed in the group of patients with OSAHS (*N* = 21).

TABLE 5 Evaluation by IOPI test in the healthy control, PS and OSAHS groups before performing oropharyngeal exercises

	IOPI-t	IOPI-l	p^a
Control group (*N* = 20)	66 ± 18.2	42 ± 9.3	Control vs PS group; <0.05*
PS (*N* = 22)	42 ± 16.7	29 ± 13.4	Control vs OSAHS group; 0.03*
OSAHS (*N* = 21)	31 ± 19.5	21 ± 8.8	PS group vs OSAHS group; 0.06

Abbreviations: IOPI, Iowa oral performance instrument; l, lips; OSAHS, sleep apnoea-hypopnea syndrome; PS, primary snoring; t, tongue.

[a]Mann-Whitney test.

*represents the significant results in the statistical analysis for a *p* value <0.05.

to complete the exercises. The app was successful in this very homogeneous and well-selected population cohort (middle-aged/elderly white men). Komoda et al.[28] investigated the effect of repeated tongue lift training (TLT) on the excitability of the corticomotor representation of the human tongue and jaw musculature. Sixteen participants underwent transcranial magnetic stimulation and electromyographic recordings of motor-evoked potentials. Their results suggest that repeated and standardised application of TLT triggers significant neuroplastic changes in corticomotor representation, of not only the tongue musculature but also the jaw-closing muscles. These results have implications regarding functional aspects of oral rehabilitation and could lead to an improvement in treatment procedures.

We used the IOPI to measure the tongue's motor responses after patients performed the oropharyngeal exercises. We found significant changes in these responses after the therapy. Few studies have focused on using IOPI to measure tongue and lip tone in patients with OSAHS. A systematic review published in 2013 by Adams et al.[29] found that IOPI has been used mainly to measure tongue strength (38 studies) and resistance (15 studies). Eighty per cent of population-based studies have focused on speech or swallowing pathologies.[20,30-32] Here, we wanted to measure this impairment in orofacial strength and to determine whether it could be corrected using an MT approach. We found that using the Airway Gym® app resulted in values within the normal ranges for the IOPI in patients with OSAHS and those with PS, which were much worse prior to treatment.[17] A recently published study on the use of IOPI in patients with OSAHS by O'Connor-Reina et al.[14] used tongue peak scores as a useful tool in the topographic diagnosis of airway collapse in patients with OSAHS. Another recent article is the study by Arakawa et al.[33] in which they evaluate the comparability between the quantitative clinical evaluation tests of oral function. To do this, they used pairs of instruments to measure maximum tongue pressure (MTP), chewing function (CF) and maximum lip force (MLF) in 26 healthy subjects.

The values with the MTP measured with the IOPI or with the JMS (TPM-01 device, JMS Co., Ltd.), can be comparable to each other ($r = 0.39$; $p < .049$). MLF values measured by different plates [smaller plate (29 × 17 mm) (left) and larger plate (Ulmer Modell Oral Screen; dimensions 55 × 24 mm)] and the CF measured by two different chewing gums (Hue-Chek and Vivident) cannot be directly compared to each other, CF ($r = 0.32$; $p = 0.113$) and MLF ($r = 0.31$; $p = 0.115$).

Studies have reported 15% higher type IIA muscle fibres in various airway muscles, such as the uvula, genioglossus, middle pharyngeal constrictor and palatopharyngeal muscles, in patients with OSAHS compared with healthy subjects. This could be relevant because these are fast-twitch fibres that use both aerobic and anaerobic metabolism but are less fatigue-resistant than other types of muscle fibres.[4] We wonder whether the muscle fibre distribution might have changed following exercises with this app in our patients. Further investigations should be performed to assure this point.

Several works have demonstrated the effectiveness of the Airway Gym® app in the therapy of patients with SDB.[17-19,34] One of the main weaknesses of the studies published on MT is that they used different groups of exercises to treat the patients. This app offers the same group of exercises that allow an objective evaluation of the efficacy of the therapy. Another limitation of conventional MT is its low adherence by participants.[35] However, in our series, we achieved successful compliance rates of 84% in groups of OSAHS the and 82% in the PS group following MT. However, we wonder, if the results in long-term follow-up therapy will be the same. As has been suggested previously,[17] we believe that the main success of this app is that it provides proprioceptive rehabilitation based on visual, acoustical and tactile feedback.

As O'Connor-Reina et al. reported,[17] the exercises performed using Airway Gym showed effectiveness in severe OSAHS, but they were not designed to treat conventional myofunctional disorders. Under our experience, we proposed a new name for oropharyngeal exercises when treated SDB as 'sensorimotor muscle rehabilitation' (SMR) as some of SDB patients had mainly hypotony and sensorimotor deficits. We recommend the use of myofunctional therapy exclusively when these exercises are designed to improve tongue position, improve lip seal and enhance nasal breathing.

4.1 | Limitations

Our results should be interpreted with caution because of the limited number of participants, and because our investigation was non-randomised. Other limitations were that we did not verify improvement in the muscular response capacity using PSG as the objective of this work was to evaluate the sensorimotor improvement after the use of this app in patients with SDB. We also need to question whether the improvements in lingual gnosis resulted from the loss of fat in the tongue or from an improvement in muscle tone, although we found no significant change in neck circumference, which suggests that this app works by increasing muscle tone and sensorimotor deficits. Using magnetic resonance imaging (MRI) of the neck, Marin et al.[36] demonstrated that the total volume of fat in the upper airway is more significant in patients with OSAHS than that in healthy subjects. This change is accompanied by decreased airway diameter, an increase in the resistance and greater collapsibility. A recent study using volumetric MRI showed that tongue and lateral wall volumes are independent risk factors for OSAHS; weight loss decreases tongue fat, and this is a mediator of the improvement in AHI.[37] We suggest that one possible investigation in future would be to perform imaging studies in such patients undertaking oropharyngeal exercises to evaluate the possible reduction of fat in the airway as a means of reducing the obstruction. Another limitation of this study is the concomitant use of the CPAP in the OSAHS group during the performance of the exercises, as it might impact the results of the investigation. In O'Connor-Reina et al,[17] in their RCT based on two groups with severe OSAHS, one group without any therapy and other only using this app, there were no significant changes in the control group in AHI and ODI parameters, and these changes were found in the study group. We are starting a new RCT just with severe OSAHS patients evaluating sensorimotor answers and respiratory parameters with this app compared with sham therapy and no CPAP as concomitant therapy.

5 | CONCLUSIONS

This is the first report of sensorimotor monitoring in adult patients with sleep disorders such as PS and OSAHS after completion of MT. The exercises used by the Airway Gym® app appear to provide a therapeutic tool for improving motor tongue function and correcting lingual stereognosis. These results might reflect the changes induced by the continuous brain and sound stimulation required to localise the tongue while using the app. Thus, we offer a new well-tested and simple protocol in the evaluation of sensorimotor impairments of patients with SDB with potential therapeutic implications.

ACKNOWLEDGEMENTS

We acknowledge the work of Professor O'Connor-Reina who designed and produced the Airway Gym® app for his patients and whose work was central to this research.

CONFLICT OF INTEREST

None declared.

AUTHOR'S CONTRIBUTIONS

The patients were under the care of LRA and JML. LRA, JML and RRF conceived and design the analysis. LRA and PB contributed data and analysis tools. LRA and JPS performed the analysis. LRA, JML and GP wrote the report. FJG, PB, RRF and GP supervised.

PEER REVIEW

The peer review history for this article is available at https://publons.com/publon/10.1111/joor.13247.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ORCID

Laura Rodríguez-Alcalá https://orcid.org/0000-0002-5148-5111

REFERENCES

1. Peppard PE, Young T, Barnet JH, Palta M, Hagen EW, Hla KM. Increased prevalence of sleep-disordered breathing in adults. *Am J Epidemiol*. 2013;177(9):1006-1014.
2. Malhotra A, Fogel RB, Edwards JK, Shea SA, White DP. Local mechanisms drive genioglossus activation in obstructive sleep apnea. *Am J Respir Crit Care Med*. 2000;161(5):1746-1749.
3. White DP. Pathogenesis of obstructive and central sleep apnea. *Am J Respir Crit Care Med*. 2005;172(11):1363-1370.
4. Remmers JE, deGroot WJ, Sauerland EK, Anch AM. Pathogenesis of upper airway occlusion during sleep. *J Appl Physiol*. 1978;44(6):931-938.
5. Guilleminault C, Huang YS, Quo S. Apraxia in children and adults with obstructive sleep apnea syndrome. *Sleep*. 2019;42(12):zsz168.
6. Hanning CD. Laryngeal and velopharyngeal sensory impairment in obstructive sleep apnea. *Sleep*. 2005;28(10):1335.
7. De Felício CM, da Silva Dias FV, Trawitzki LV. Obstructive sleep apnea: focus on myofunctional therapy. *Nat Sci Sleep*. 2018;10:271-286.
8. Guimarães KC, Drager LF, Genta PR, Marcondes BF, Lorenzi-Filho G. Effects of oropharyngeal exercises on patients with moderate obstructive sleep apnea syndrome. *Am J Respir Crit Care Med*. 2009;179(10):962-966.
9. Ieto V, Kayamori F, Montes MI, et al. Effects of oropharyngeal exercises on snoring: a randomized trial. *Chest*. 2015;148(3):683-691.
10. Diaféria G, Santos-Silva R, Truksinas E, et al. Myofunctional therapy improves adherence to continuous positive airway pressure treatment. *Sleep Breath*. 2017;21(2):387-395.
11. Camacho M, Guilleminault C, Wei JM, et al. Oropharyngeal and tongue exercises(myofunctional therapy) for snoring: a systematic review and meta-analysis. *Eur Arch Otorhinolaryngol*. 2018;275(4):849-855.
12. Villa MP, Evangelisti M, Martella S, Barreto M, Del Pozzo M. Can myofunctional therapy increase tongue tone and reduce symptoms in children with sleep-disordered breathing? *Sleep Breath*. 2017;21(4):1025-1032.
13. De Felício CM, da Silva Dias FV, Folha GA, et al. Orofacial motor functions in pediatric obstructive sleep apnea and implications for myofunctional therapy. *Int J Pediatr Otorhinolaryngol*. 2016;90:5-11.
14. O'Connor-Reina C, Plaza G, Garcia-Iriarte MT, et al. Tongue peak pressure a tool to aid in the identification of obstruction sites in patients with obstructive sleep apnea/hypopnea syndrome. *Sleep Breath*. 2020;24:281-286.
15. Buterbaugh J, Wynstra C, Provencio N, et al. Cerebrovascular reactivity in young subjects with sleep apnea. *Sleep*. 2015;38(2):241-250.
16. Aman JE, Elangovan N, Yeh IL, Konczak J. The effectiveness of proprioceptive training for improving motor function: a systematic review. *Front Hum Neurosci*. 2015;8:1075.
17. O'Connor-Reina C, Ignacio-Garcia JM, Rodriguez-Ruiz E, et al. Myofunctional therapy app for severe apnea–hypopnea sleep obstructive syndrome: a pilot randomized controlled trial. *JMIR Mhealth Uhealth*. 2020: 23123(in press). 10.2196/23123. URL: https://preprints.jmir.org/preprint/23123
18. O'Connor-Reina C, García-Iriarte MT, Casado-Morente JC, et al. New app "Apnea Bye" increases adherence in myofunctional therapy to treat sleep disordered breathing. *Otolaryngol Head Neck Surg*. 2018;159:32
19. O'Connor-Reina C, Plaza G, Ignacio-García JM, et al. New mHealth application software based on myofunctional therapy applied to sleep-disordered breathing in non-compliant subjects. *Sleep Sci Pract*. 2020;4(3):1-10.
20. Steele CM, Bayley MT, Peladeau-Pigeon M, et al. A randomized trial comparing two tongue-pressure resistance training protocols for post-stroke dysphagia. *Dysphagia*. 2016;31(3):452-461.
21. Marchesan I. Lingual frenulum protocol. *Int J Orofac Myol*. 2012;38:89-103.
22. Beamont KG, Gokhale A, Heaphy N. Temporomandibular Disorder: a practical guide por dental practitioners in diagnosis and management. *Aust Dent J*. 2020. 10.1111/adj.12785.
23. Chiner E, Arriero JM, Signes-Costa J, Marco J, Fuentes I. Validation of the Spanish version of the Epworth Sleepiness Scale

in patients with a sleep apnea syndrome. *Arch Bronconeumol*. 1999;35(9):422-427.
24. Larrosa F, Roura J, Dura MJ, Guirao M, Alberti A, Alobid I. Adaptation and validation of the Spanish version of the Nasal Obstruction Symptom Evaluation(NOSE) scale. *Rhinology*. 2015;53(2):176-180.
25. Robin DA, Luschei ES. *IOPI: Iowa Oral Performance Instrument Reference Manual*. Breakthrough; 1992.
26. Berry RB, Gamaldo CE, Harding SM, et al. AASM Scoring Manual Version 2.2 Updates: New chapters for scoring infant sleep staging and home sleep apnea testing. *J Clin Sleep Med*. 2015;11(11):1253-1254.
27. Eckert DJ. Phenotypic approaches to obstructive sleep apnoea – new pathways for targeted therapy. *Sleep Med Rev*. 2018;37:45-59.
28. Komoda Y, Iida T, Kothari M, et al. Repeated tongue lift movement induces neuroplasticity in corticomotor control of tongue and jaw muscles in humans. *Brain Res*. 2015;1627:70-79.
29. Adams V, Mathisen B, Baines S, Lazarus C, Callister R. A systematic review and meta-analysis of measurements of tongue and hand strength and endurance using the Iowa Oral Performance Instrument (IOPI). *Dysphagia*. 2013;28(3):350-369.
30. Clark HM, Solomon NP. Age and sex differences in orofacial strength. *Dysphagia*. 2012;27(1):2-9.
31. Youmans SR, Stierwalt JAG. Measures of tongue function related to swallowing. *Dysphagia*. 2006;21(2):102-111.
32. Adams V, Mathisen B, Baines S, Lazarus C, Callister R. Reliability of measurements of tongue and hand strength and endurance using the Iowa Oral Performance Instrument with healthy adults. *Dysphagia*. 2014;29(1):83-95.
33. Arakawa I, Abou-Ayash S, Genton L. Reliability and comparability of methods for assessing oral function: Chewing, tongue pressure and lip force. *J Oral Rehabil*. 2020;47:862-871.
34. O'Connor-Reina C, Plaza Mayor G, Ignacio-Garcia JM, et al. Floppy closing door epiglottis treated successfully with an Mhealth application based on myofunctional therapy: a case report. *Case Rep Otolaryngol*. 2019;2019:4157898.
35. Mario Diaz S, Ana Salazar C, Felipe Bravoi G, Ocampo-Garcés A. Tratamiento del síndrome de apneas e hipopneas obstructivas del sueño con terapia miofuncional orofaríngea: Experiencia en hospital público de Chile. *Rev Otorrinolaringol Cir Cabeza Cuello*. 2019;79(4):395-403.
36. Marín JM, Kimoff JR, Cosio MG. Obstructive sleep apnea syndrome and its treatment by continuous nasal positive air pressure. *Arch Bronconeumol*. 1990;26(6):255-261.
37. Wang SH, Keenan BT, Wiemken A, et al. Effect of weight loss on upper airway anatomy and the apnea–hypopnea index. The importance of tongue fat. *Am J Respir Crit Care Med*. 2020;201(6):718-727.

SUPPORTING INFORMATION

Additional supporting information may be found in the online version of the article at the publisher's website.

How to cite this article: Rodríguez-Alcalá L, Martínez JM-L, Baptista P, et al. Sensorimotor tongue evaluation and rehabilitation in patients with sleep-disordered breathing: a novel approach. *J Oral Rehabil*. 2021;48:1363–1372. https://doi.org/10.1111/joor.13247