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HEAD AND NECK

Moderate-to-severe obstructive sleep apnea syndrome is associated with altered tongue motion during wakefulness

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Abstract

Purpose Impairment of genioglossus control is a frequent "non-anatomical" cause of obstructive sleep apnea syndrome (OSAS) in non- or mildly obese patients. Although wake-related compensatory mechanisms prevent the occurrence of obstructive events, the genioglossus control is often impaired during wakefulness. We hypothesized that the lingual motion would be altered during wakefulness in this population in patients with moderate-to-severe OSAS.

Methods We included non- or mildly obese participants with suspected OSAS. They underwent a Bucco-Linguo-Facial Motor Skills assessment using the MBLF ("Motricité Bucco-Linguo-Faciale"), which includes an evaluation of 13 movements of the tongue. This was followed by a night-attended polysomnography. We compared patients with moderate-to-severe OSAS (apnea–hypopnea index (AHI) ≥ 15/h; $n = 15$) to patients without or with mild OSAS (AHI < 15/h; $n = 24$).

Results MBLF total and "tongue" sub-scores were lower in patients with moderate-to-severe OSAS: total z-score −0.78 [−1.31; 0.103] versus 0.20 [−0.26; 0.31], $p = 0.0011$; "tongue" z-sub-score (−0.63 [−1.83; 0.41] versus 0.35 [0.26; 0.48], $p = 0.014$). There was a significant age-adjusted correlation between the "tongue" sub-score and AHI. The logistic regression model for the prediction of moderate-to-severe OSAS gave area under the curve ratio of 88.2% for MBLF score plus age.

Conclusions Myofunctional activity of the tongue is impaired during wakefulness in non- or mildly obese patients with moderate-to-severe OSAS. This study supports the lingual myofunctional assessment using the MBLF in screening of moderate-to-severe OSAS. This simple tool could help clinicians to select patients with suspected moderate-to-severe OSAS for polysomnography.

Keywords Obstructive sleep apnea syndrome · Orofacial myofunctional activity · Tongue · Voice · MBLF

Abbreviations

AHI	Apnea–hypopnea index
AUCs	Area under the receiver-operating characteristic (ROC) curve
BMI	Body mass index
ESS	Epworth Sleepiness Scale
HNR	Harmonic-to-noise ratio
MBLF	Motricité Bucco-Linguo-Faciale (oral-linguo-facial motor skills)
MPT	Maximum phonation time
OSAS	Obstructive sleep apnea syndrome

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PSG Polysomnography
PSQI Pittsburgh Sleep Quality Index
VHI Voice Handicap Index

Introduction

Obstructive sleep apnea hypopnea syndrome (OSAS) is a common disorder characterized by repetitive, brief obstructions of upper airways during sleep [1]. The obstructions cause intermittent hypoxia and hypercapnia, sleep fragmentation, and increased respiratory efforts, which in turns promote, in patients with moderate-to-severe OSAS, cardiovascular, metabolic, and neurocognitive comorbidities [2]. Patients with moderate-to-severe OSAS should be given priority for polysomnography and treatment. In patients without severe obesity referred for suspected OSAS, visual examination of upper airways may be poorly informative as a screening to select patients with suspected moderate-to-severe OSAS, for polysomnography. Indeed, upper airway obstruction during sleep can be caused by anatomical and/or functional factors, leading to various pathophysiology phenotypes [3]. On the one hand, the upper airway size and shape may be reduced by narrow craniofacial structures, as well as fat deposits in pharyngeal walls. On the other hand, upper airway (which is a muscular and membranous extrathoracic tube) may have a normal diameter, but collapse during sleep when inspiration generates a negative pressure [3]. Indeed, in humans, inspiration exposes upper airway to a negative pressure, which tends to collapse it [4]. The upper airway stability during the respiratory cycle is then the result of both tonic [5] and phasic activity of pharyngeal dilatator muscles, including tongue muscles [6]. This illustrates the respiratory function of the tongue, the genioglossus (more precisely its posterior part) being one of the main tongue muscles involved in maintaining the pharyngeal patency during inspiration [5]. During normal breathing, the genioglossus maintains upper airway patency throughout the complete respiratory cycle. It contracts before the diaphragm (i.e., before inspiration), induces a mild anterior displacement of the tongue that lasts the entire time of the inspiration [6], opposes the collapse generated by the diaphragmatic contraction, and helps to stabilize the pharynx [4, 7]. Impairment of pharyngeal dilator muscles control is a frequent functional (i.e., "non-anatomical") cause of upper airway collapse during sleep [3], in non-obese or mildly obese patients with OSAS [8]. In this population, the genioglossus control is also impaired during wakefulness [9], while wake-related compensatory mechanisms explain the absence of obstructive events [10–12]. Indeed, while awake, it has been reported in patients with OSAS, compared to control, a less stiff tongue [13], a pathological coordination of lingual muscles which correlates with OSAS severity [14], and greater motion of the genioglossus during inspiration [15]. In addition, an impaired tongue strength in OSAS patients compared to controls was reported [16, 17] as well as a significant correlation between the tongue pressure and the upper airways size during drug-induced sleep endoscopy [16]. We then hypothesized that the orofacial myofunctional activity and particularly the tongue motion would be impaired in moderate-to-severe OSAS patients, compared to subjects without or with mild OSAS. We carefully assessed the orofacial myofunctional activity in patients with moderate-to-severe OSAS (AHI $\geq$ 15) and in patients with AHI < 15 as a control group, using the MBLF (Motricité Bucco-Linguo-Faciale) test, which was recently validated in healthy subjects and patients with facial palsy [18]. This test consists of a rapid clinical evaluation of bucco-linguo-facial motor skills and includes an evaluation of 13 movements of the tongue. We then evaluated if the "tongue" sub-score of the MBLF was predictive of moderate-to-severe OSAS. In addition, we studied whether any impairment in orofacial myofunctional activity would correlate with voice changes.

Methods

Participants

Participants were recruited in the Sleep Disorders Unit of Pitié-Salpêtrière University Hospital (Paris, France) over a period of 3 months among patients referred for an attended polysomnography. None had temporomandibular limitation, tongue tie, or movement restriction of the tongue due to the lingual frenulum, severe nasal obstruction, or prior surgery in the upper airway. Inclusion criteria were: adults (age $\geq$ 18 years); suspicion of OSAS; a body mass index (BMI) < 35 kg/m^2; and willing to take part in the study. Non-inclusion criteria were: previously diagnosed OSAS; concurrent neurological disorder; chronic respiratory disease other than OSAS; and language, hearing or uncorrected visual disorder. The study was performed in accordance with the ethical standards as laid down in the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards. The protocol was approved by ethics committee (Comité de Protection des Personnes Ouest II, Angers, France, 2019; number: 2018-A03250-55). All participants were informed about the study and provided their written consent. The two groups were later constituted according to an apnea–hypopnea index (AHI) below (AHI < 15 group) or equal to or greater than (AHI $\geq$ 15 group; moderate-to-severe OSAS group) 15.

Procedures

In the afternoon, participants completed the Epworth Sleepiness Scale (ESS) [19] and Pittsburgh Sleep Quality Index

(PSQI) [20]. A speech therapist evaluated their tongue mobility and voice (see below). Then, participants underwent a full-night-attended video-polysomnography (PSG) (Compumedics France SAS), including a three channel (F3/A2, C3/A2, C3/01) EEG, left and right electro-oculograms, surface electromyograms of chin and legs muscles, electrocardiogram, transcutaneous oxygen saturation, thoracic and abdominal effort detection by belts, and measure of airflow using a nasal pressure transducer and a naso-oral thermistor. The polysomnography recording was blind-scored by a single experienced sleep medicine physician, according to the American Academy of Sleep Medicine scoring rules [21]. Blinding of the speech therapist was not necessary as he did not know the result of the PSG.

Orofacial myofunctional assessment

The orofacial myofunctional activity was evaluated using the computerized version of the Motricité Bucco-Linguo-Faciale (MBLF) test (Adeprio Logiciels, Gisors, France) [18]. This test takes around 10 min. It assesses several movements of the face, the eyes, the lips, the cheeks/jaw, and the tongue, which are each scored from 0 (no contraction) to 3 (normal contraction). The maximal score is 39 for the tongue (13 tested movements), 6 for the face (2 movements), 27 for the lips (9 movements), 9 for the eyes (3 movements), and 30 for the cheeks (10 movements). The total score, which is the sum of all area scores, ranges from 0 to 111 (Table 1 and Fig. 1). As recommended, we computed age-adjusted *z*-scores which include a correction for age (pathological if ≤ -1.65, i.e., in the 5% left tail) [18]. We characterized the anatomy of the oral cavity using the Mallampati score [22], assessed by asking the participants to open their mouth and stick out their tongue. This score includes four categories: class I—the uvula and the tonsils are visible; class II—the uvula is partially visible; class III—the soft palate is visible but not the uvula; class IV—only the bony palate is visible.

Voice assessment

The voice was recorded using a microphone Yeti Pro (Blue Microphones, Westlake Village, California, United States). The acoustic parameters of the voice were analyzed using Praat computer software (retrieved 23 January 2022 https://www.praat.org). The patients were sitting in front of the microphone positioned 20 cm away from their mouth and were asked to articulate the vowel /a/ for as long as possible. Three /a/'s were recorded and the longest was used to determine the maximum phonation time (MPT), which is the maximum time the patient can sustain the vowel, in seconds (standard ≥ 15 s, weak $9 \leq x < 15$ s, pathological < 9 s). The fundamental frequency (F0) is the vibration rate of the vocal folds (standard for men are around 110 Hz, for women around 220 Hz). The jitter is the cycle-to-cycle variation of the fundamental frequency, it reflects the inability of the vocal folds to support periodic vibration, and the presence of turbulence noise in the voice signal (standard $< 1.04\%$) [23]. The shimmer is the fluctuation of voice amplitude, representing the period-to-period variability of voice (standard $< 3.81\%$) [23]. Harmonic-to-noise ratio (HNR) is the amplitude of tonal components relative to noise (standard ≥ 20 decibels). In addition, we performed a subjective assessment of the voice using the Voice Handicap Index (VHI) self-assessment questionnaire which comprises 30 items and assesses the emotional, physical, and functional characteristics of the voice leading to a score ranging from 0 to 120 [24–28]: the higher the score, the higher the perceived handicap.

Statistical analyses

The statistical analyses were performed using MATLAB (R2021a). As most variables did not follow a normal distribution, we used Wilcoxon's non-parametric test to compare continuous variables between both groups. Fisher's exact test was used to compare proportions. The correlations of the MBLF scores with AHI were evaluated using Spearman's non-parametric coefficient on all patients. These correlations were computed unadjusted and adjusted for age. A logistic regression model was used to predict OSAS using MBLF alone, MBLF + age and MBLF + age + BMI. For all tests, a p value was considered significant if < 0.05.

Results

During the 3-month study period, 190 patients were referred for a one-night polysomnography for suspected OSAS among whom 39 were included. After polysomnography scoring, 15 patients had a moderate-to-severe OSAS with an AHI ≥ 15/h (AHI ≥ 15 group) and 24 patients had an AHI < 15/h (AHI < 15 group). The flowchart is depicted in Fig. 2.

Baseline characteristics and polysomnography

Patients of both groups were comparable for gender and BMI, but patients with AHI ≥ 15 were older (Table 2). All participants snored and a similar proportion of participants reported high intensity snoring, but more from the AHI ≥ 15 group snored every night. There was no difference between groups concerning other OSAS symptoms, ESS, and sleep quality (PSQI). By definition, patients with AHI ≥ 15 had higher AHI than patients with AHI < 15. In addition, the percentage of N3 sleep was lower in patients with AHI ≥ 15, while the other sleep measures were similar in the two groups.

Table 1 MBLF protocol

Facial areas	Oral motor tasks	Muscles	0	1	2	3	Score
Face	Symmetry at rest						
	Symmetry when smiling						
	/6						
Eyes	Close your eyes	Orbicularis oculi					
	Raise your eyebrows	Occipito-frontalis					
	Frown	Corrugator supercilii					
	/9						
Lips	Pinch your lips	Compressor/buccinator					
	Stretch your lips	Zygomaticus/risorius					
	Keep your lips closed strongly	Orbicularis oris/masseter					
	Open mouth smile	Zygomaticus/risorius					
	Show the upper teeth	Levator labii superioris					
	Show the lower teeth	Mentalis					
	Say "u"	Orbicularis oris					
	Whistle	Orbicularis oris					
	Blow	Orbicularis oris					
	/27						
Cheeks and mandibles	Open your mouth	Buccinator/orbicularis oris					
	Close your mouth	Masseter/orbicularis oris					
	Puff off the cheeks	Buccinator/orbicularis oris					
	Puff left cheek	Buccinator/orbicularis oris					
	Puff right cheek	Buccinator/orbicularis oris					
	Pass the air from one cheek to another	Buccinator/orbicularis oris					
	Suck in the cheeks	Buccinator/orbicularis oris					
	Left jaw open mouth	Pterygoid					
	Right jaw open mouth	Pterygoid					
	Chew closed mouth						
	/30						
Tongue	Stick the tongue out	Genioglossus/transverse					
	Bring in the tongue	Hyoglossus/superior longitudinal					
	Put the tongue to the right corner of the mouth	Pharyngoglossus					
	Put the tongue to the left corner of the mouth	Pharyngoglossus					
	Put it on top	Superior longitudinal					
	Put it down	Superior longitudinal					
	Put your tongue on your teeth	Styloglossus/hyoglossus					
	Move the tongue inside the right cheek						
	Move the tongue inside the left cheek						
	Raise the tip in the mouth	Pharyngoglossus					
	Raise the tip out of the mouth	Styloglossus					
	Click of disagreement	Styloglossus					
	Rhythm of galloping horse	Styloglossus					
	/39						

MBLF: Motricité Bucco-Linguo-Faciale (oral-linguo-facial motor skills); scoring of face symmetry: 0 = severe/complete asymmetry; 1 = significant/moderate asymmetry; 2 = mild asymmetry; 3 = complete symmetry; scoring of muscle contraction Rating: 0 = no contraction; 1 = initiated movement; 2 = almost complete movement; 3 = normal contraction; total score /39

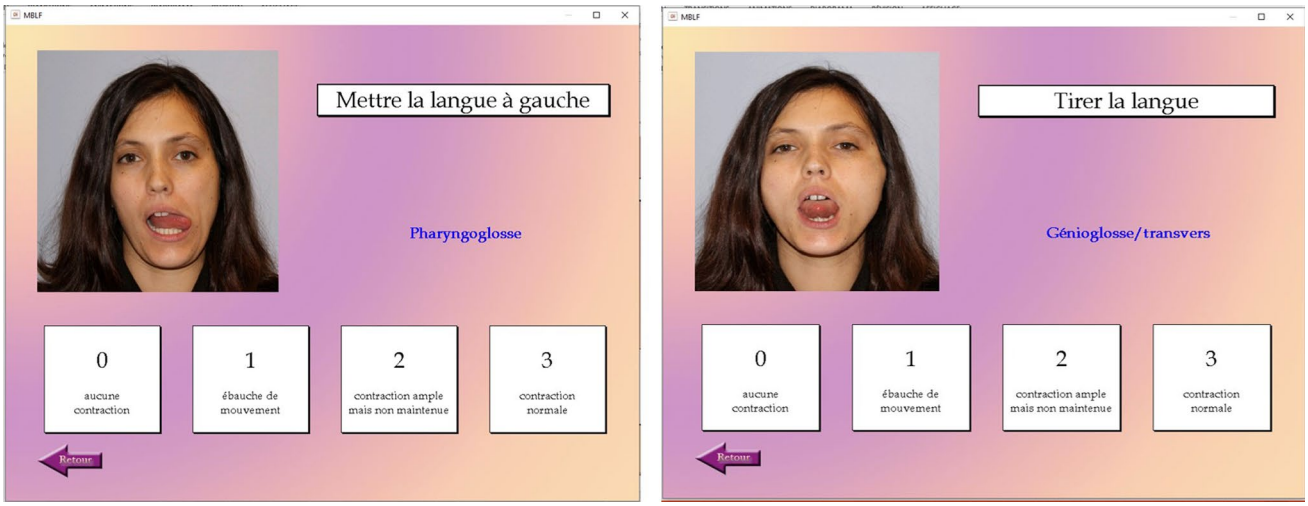

Fig. 1 Computerized version of the MBLF test. Scoring of orofacial myofunctional activity using the computerized version of the Motricité Bucco-Linguo-Faciale (MBLF) (Adeprio Logiciels, Gisors, France). Examples of two tongue movements; left: "Put the tongue to the left corner of the mouth", right: "stick the tongue out": 0 = no contraction; 1 = initiated movement; 2 = almost complete movement; 3 = normal contraction. Written consent was obtained for publication of these images

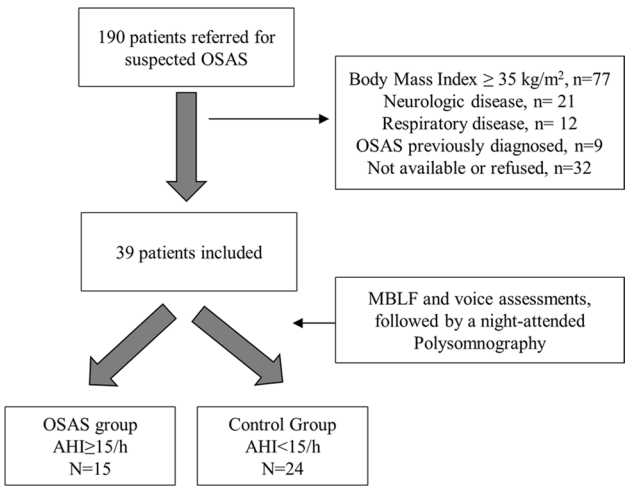

Fig. 2 Study flowchart

Orofacial myofunctional assessment

The MBLF total score and the "tongue" and "cheeks" MBLF sub-scores were lower in the moderate-to-severe AHI ≥ 15 group (Table 3). There was a significant non-adjusted correlation between these three scores and AHI, which was evident after adjustment for age only for total score and "tongue" sub-score. More patients had a MBLF "tongue" z-sub-score < 0 in the group with AHI ≥ 15 compared to the group with AHI < 15 (10/15 patients (66.7%) versus 3/24 patients (12.5%); $p = 0.001$). For the MBLF "cheek" z-sub-score, 6/15 patients (40.0%) had a z-sub-score < 0 in the group with AHI ≥ 15 versus 3/24 patients (12.5%) in the group with AHI < 15 ($p = 0.063$). Similarly, patients with AHI ≥ 15 had lower total MBLF age-adjusted z-scores and "tongue" z-sub-score than patients with AHI < 15 (AHI ≥ 15 versus AHI < 15: MBLF total z-score −0.78 [−1.31; 0.103] versus 0.20 [−0.26; 0.31], $p = 0.0011$; "tongue" z-sub-score (−0.63 [−1.83; 0.41] versus 0.35 [0.26; 0.48], $p = 0.014$) (Fig. 3). The logistic regression model for the prediction of OSAS gave area under the ROC curve (AUCs) of 76.9% ($p = 0.008$) for MBLF alone, 88.2% ($p = 0.038$) for MBLF plus age, and 88.3% ($p = 0.034$) for MBLF plus age and BMI (Fig. 4).

Voice assessment

No difference was observed between groups (Table 4). The Maximum Phonation Time, the Fundamental frequency, the Jitter and the Voice handicap Index were in the normal range for patients with AHI ≥ 15 and patients with AHI < 15, but almost all participants had abnormal shimmer and harmonic-to-noise ratio.

Discussion

This study shows that awake non-obese or non-severely obese patients with moderate-to-severe OSAS have an impaired myofunctional activity of the tongue when compared with non- or mild OSAS subjects. This lingual dysfunction is not linked to an alteration of the phonatory system.

Previous studies reported an orofacial myofunctional impairment in awake patients with OSAS [25–29], including impaired lingual praxis [29], and a tongue dysfunction

Table 2 Baseline characteristics and polysomnography

	Patients with AHI < 15, *N* = 24	Patients with AHI ≥ 15, *N* = 15	*p*
Baseline characteristics			
Gender (male/female; *n*)	11/13	10/5	0.323
Age (years)	48 [31; 57]	63 [55; 68]	0.0008
Body mass index (kg/m^2)	26.5 [23.7; 30.0]	27.1 [25.0; 30.9]	0.479
Mallampati score, I/II/III/IV	0/2/3/10	5/4/7/8	0.146
Snoring every nights (% subjects)	54	93	0.013
Snoring with high intensity (% subjects)	50	60	0.742
Dry mouth (% subjects)	33	27	0.734
Night voiding (% subjects)	46	47	1
Night voiding (n/night)	0.0 [0.0; 2.0]	0.0 [0.0; 2.0]	0.537
Morning headache (% subjects)	38	33	1
Tiredness (% subjects)	79	47	0.079
Subjective somnolence (% subjects)	54	47	0.748
Epworth sleepiness score (0–24)	8.5 [6.0; 12.5]	8.0 [3.3; 11.8]	0.271
PSQI (0–21)	7.5 [5.0; 10.5]	7.0 [3.8; 10.8]	0.622
PSQI (% subjects with a score > 5)	67	73	0.734
Polysomnography			
Sleep onset latency (min)	12.8 [7.3; 30.0]	14.5 [9.8; 34.9]	0.419
Total sleep time (TST, in min)	415.8 [376.5; 472.5]	391.0 [323.1; 476.1]	0.748
Sleep efficiency (%)	87.4 [79.5; 93.1]	82.7 [69.5; 93.5]	0.593
N1 Sleep (% TST)	2.7 [1.5; 5.6]	4.0 [1.5; 12.6]	0.602
N2 Sleep (% TST)	53.2 [42.4; 59.5]	57.0 [49.3; 67.8]	0.153
N3 Sleep (% TST)	21.0 [17.0; 27.6]	16.3 [10.3; 21.7]	0.045
REM sleep (% TST)	21.7 [18.8; 24.8]	18.2 [13.6; 23.8]	0.299
Arousal Index (n/h)	7.2 [4.0; 11.6]	23.8 [12.9; 32.1]	< 0.0001
Apnea–Hypopnea Index (n/h)	1.4 [0.1; 6.0]	30.0 [19.0; 64.4]	< 0.0001
Apnea index (n/h)	0.0 [0.0; 0.6]	8.6 [4.1; 30.1]	< 0.0001
Oxygen Desaturation Index, 3% (n/h)	1.7 [0.7; 4.6]	21.2 [13.3; 49.8]	< 0.0001
SpO^2 < 90% (% of TST)	0.0 [0.0; 0.5]	6.3 [0.8; 9.8]	< 0.0001
Periodic limb movements (n/h)	0.8 [0.0; 10.4]	1.0 [0.0; 11.9]	0.808

Results are expressed as median and interquartile interval or percentage. *p* value, Wilcoxon test for continuous variables or Fisher exact test for proportions

OSAS Obstructive Sleep Apnea Syndrome, *PSQI* Pittsburgh Sleep Quality Index

which was correlated to OSAS severity [28], and alterations in cheek, lip, and jaw mobility [27]. The scope of the conclusions of these studies is, however, limited by heterogeneity of patients (e.g., including severely obese plus non-obese patients) [25–28], or absence of control subjects [28]. Consequently, deciphering functional from anatomical causes of lingual dysfunction (e.g., increased tongue volume caused by fat infiltrates) is difficult. We aimed at circumventing these limitations by including only non- or mildly obese patients for several reasons. First, obesity itself induces fat accumulation in the tongue [30] and is associated with smaller upper airway size [31], changes in oral motor function [32], greater anterior motion of the genioglossus during inspiration [31], and enhanced upper airway dilator muscle responses [33]. Second, patients with OSAS and moderate-to-severe obesity have a reduced upper airway volume [34] and a larger tongue compared to non apneic with moderate-to-severe obesity [35]. Third, the upper airway obstruction during sleep is mainly driven by anatomy in patients with obesity and moderate-to-severe OSAS [3], while non-anatomical factors, including impairment of the genioglossus control, predominate in non- or mildly obese patients with OSAS [3, 8]. Therefore, by including patients with a BMI < 35 kg/m^2, we avoided the confounding factor of moderate-to-severe obesity, for the assessment of orofacial myofunctional impairment in OSAS. We could focus on a homogeneous population, with supposedly predominant non-anatomical factors of upper airway obstruction. Given that one of the most frequent non-anatomical factors is an ineffective upper airway

Table 3 Oro-myo-functional assessment: MBLF scores

	Patients with AHI < 15, *N* = 24	Patients with AHI ≥ 15, *N* = 15	pMW	Spearman correlations with AHI	
				Unadjusted	Adjusted for age
Total (/111)	110 [109; 111]	104 [101; 110]	0.00053*	$\rho = -0.47$ $p_s = 0.0028$*	$\rho = -0.35$ $p_s = 0.031$*
Tongue (/39)	39 [39; 39]	38 [37; 39]	0.0015*	$\rho = -0.56$ $p_s = 0.00020$*	$\rho = -0.44$ $p_s = 0.0053$*
Face (/6)	6 [6; 6]	6 [6; 6]	1.000	$\rho = 0.17$ $p_s = 0.31$	$\rho = 0.28$ $p_s = 0.089$
Lips (/27)	27 [26; 27]	26 [25; 27]	0.162	$\rho = -0.11$ $p_s = 0.52$	$\rho = -0.041$ $p_s = 0.81$
Eyes (/9)	9 [9; 9]	9 [9; 9]	0.517	$\rho = -0.11$ $p_s = 0.49$	$\rho = -0.13$ $p_s = 0.42$
Cheeks (/30)	30 [30; 30]	26 [24; 30]	0.0094*	$\rho = -0.40$ $p_s = 0.011$*	$\rho = -0.29$ $p_s = 0.080$

Results are median and interquartile interval; pMW = comparison patients with AHI < 15/patients and with AHI ≥ 15 with Mann–Whitney test; p_s = spearman's correlation with AHI

AHI Apnea–Hypopnea Index, *MBLF* "Motricité Bucco-Linguo-Faciale" tool, *OSAS* Obstructive Sleep Apnea Syndrome, *BMI* body mass index

A star "*" indicates a significant *p* value (< 0.05)

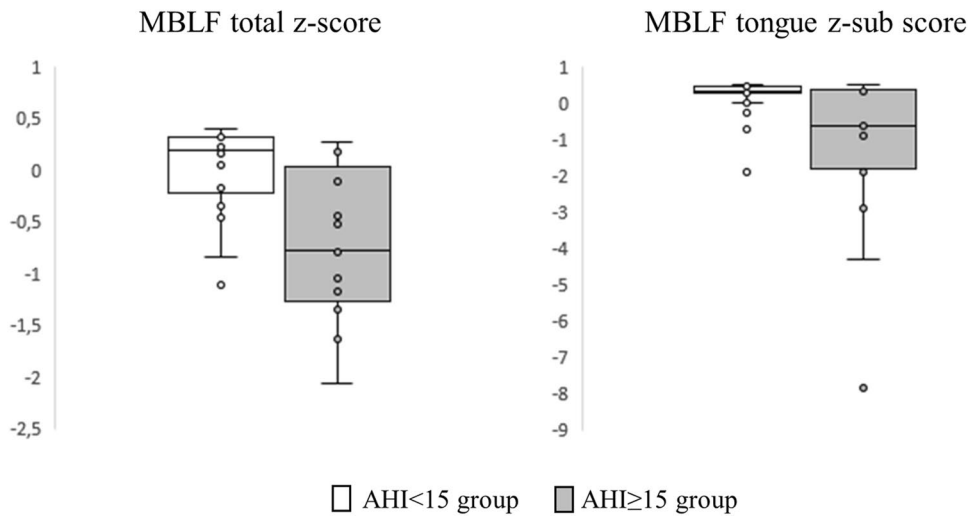

Fig. 3 MBLF age-adjusted *z*-scores. Comparison of MBLF total z-score (left) and MBLF tongue z-sub-score (right), between patients with AHI < 15 in white and patients with AHI ≥ 15 (moderate-to-severe OSAS) in gray

muscle control [3], a functional evaluation of the tongue muscles which are involved in the stability of the upper airways during breathing [4, 6] is particularly relevant. As such, our results support the lingual myofunctional evaluation using the MBLF simple tool, as a screening to select patients with suspected moderate-to-severe OSAS for polysomnography. This tool could be integrated as a complementary measurement to tongue strength [16] and more generally as a part of the myofunctional assessment in regular clinical practice [36]. In addition, our results support the use of the MBLF as a phenotypic approach to the diagnosis of moderate-to-severe OSAS [37], as well as for evaluation of lingual rehabilitation as a treatment of OSAS in selected patients [38–40].

In this study, we performed the orofacial myofunctional assessment using the MBLF (Motricité Bucco-Linguo-Faciale) which was validated in healthy subjects and in patients with facial palsy [18]. This test is easy to perform, takes around 10 min, and evaluates 13 motions of the tongue to provide a precise evaluation of lingual muscles [18]. This accurate assessment of lingual praxis in our study indicates a specific OSAS-related tongue dysfunction in awake patients. The respiratory origin of this lingual dysfunction is supported by the existence of a pathological coordination of lingual muscles during breathing, correlated to OSAS severity, that was previously reported in a study using magnetic resonance imaging [14]. In addition, an adaptation of the corticomotor conduction to the genioglossus [12] and an

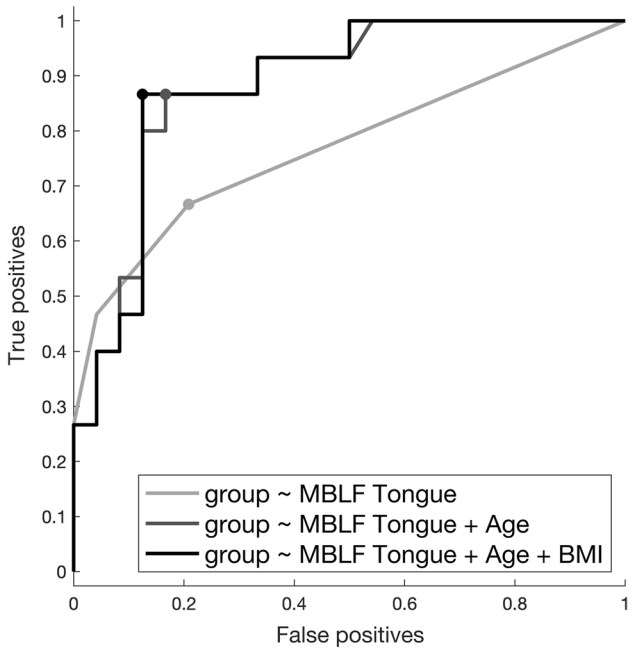

Fig. 4 Logistic regression model for prediction of OSAS. Area under the ROC curve (AUCs) of 76.9% ($p=0.008$) for MBLF alone, 88.2% ($p=0.038$) for MBLF plus age, and 88.3% ($p=0.034$) for MBLF plus age and body mass index (BMI)

Table 4 Voice assessment

	Patients with AHI < 15, $N=24$	Patients with AHI ≥ 15, $N=15$	p
Voice Handicap Index	9 [2; 16]	9 [5; 15]	0.497
Maximum phonation time (s)	12.3 [9.8; 17.6]	14.0 [12.6; 16.8]	0.521
Fundamental frequency (Hz)	145 [111; 178]	130 [121; 147]	0.644
Jitter (%)	0.4 [0.3; 1.3]	0.6 [0.3; 1.3]	0.564
Shimmer (%)	7.2 [4.8; 14.4]	7.5[6.1; 10.9]	0.638
Harmonic-to-noise ratio (db)	15.0 [11.2; 18.7]	13.5 [9.4; 17.2]	0.403

Results are median and interquartile interval. p value, Mann–Whitney test

AHI Apnea–Hypopnea Index;

increased activity of the respiratory premotor cortex [10] were reported in OSAS patients as a wake-related compensatory mechanism to prevent upper airway obstruction during wakefulness. Our results suggest that this wake-related cortical adaption is, however, not effective enough to restore a normal behavior of the tongue while awake [13]. In addition to the tongue motion evaluation, the MBLF test provides a concurrent myofunctional assessment of the face, the eyes, the lips, and the jaw. This is crucial as the genioglossus and the other extrinsic muscles of the tongue are connected to the bony and muscular structures of head and neck [41]. Consequently, though the posterior part of the genioglossus is mainly involved during inspiration, the genioglossus motion depends largely on the motion of other lingual muscles [6] and of non-lingual pharyngeal dilator muscles. In our study, we found an alteration of the cheek function that correlated with AHI, but this correlation became non-significant when adjusting for age. Larger studies are needed to investigate if this cheek alteration would be related to OSAS as previously suggested [27], or to age or to both factors.

Regarding voice assessment, patients with OSAS had normal scores for variations of F0 which tests the ability to produce a held sound and Jitter which corresponds to a variation in the voice pitch. Of note, the voice is produced primarily in the larynx and acquires its acoustic characteristics through the vocal tract. Our results suggests then the absence of a specific dysfunction of the phonatory system in OSAS. The Shimmer, i.e., the instability of the voice amplitude and the HNR, which measures the aperiodic noise present in the analyzed signal, was abnormal in patients with moderate-to-severe OSAS as expected [23, 42], but surprisingly also in patients with no or mild OSAS. As patients with AHI ≥ 15 and the majority of patients with AHI < 15 were snorers, our results support the existence of an instability of the laryngeal vibrator and a potential bad docking of the vocal cords, linked to snoring and/or mouth-breathing. Indeed, mouth-breathing and snoring would cause fluctuating modifications of the air structure of the vocal tract, and lead to irregular vibrations of the vocal cords, without vocal disorders, since the F0 remains normal. This hypothesis is supported by normalization of the voice assessment in OSAS patients after resolution of mouth-breathing under treatment with continuous positive airway pressure [23]. Our results suggest in addition that vocal assessment is not accurate for OSAS screening.

We acknowledge that our results are not generalizable to patients with OSAS and moderate-to-severe obesity and that specific studies are needed for these patients. We acknowledge that we have not collected neck and hip perimeters. We are, however, confident that in our population of non-severely-obese patients, the neck and/or hip perimeters would have low effect on the MBLF results. Our objective was to propose the MBLF as a simple tool for screening of moderate-to-severe OSAS. For this reason, we have not compared patients with OSAS to healthy participants, but we have compared patients with moderate-to-severe OSAS to patients with no or mild OSAS, in a population of patients with suspected OSAS. Our control group of patients with AHI < 15 was slightly younger. We acknowledge that this age-effect constitutes a limitation, but age was taken into account in our statistical analyses. A further limitation lies in the sample size, which restricts the power of the statistical tests: the strong association we found between cheek dysfunction and OSAS, for example, should be further investigated with a larger sample.

Conclusion

In patients with suspected OSAS, an orofacial myofunctional assessment, including a tongue evaluation, constitutes an element for moderate-to-severe OSAS screening. This study showed that the tongue motion was abnormal during wakefulness in this population with potential predominant non-anatomical functional factors of upper airway obstruction. Larger studies are needed to validate this approach, likewise for the evaluation of the lingual rehabilitation as a specific treatment.

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Data availability statement The datasets generated during and/or analysed during the current study are available from the corresponding author on reasonable request.

Declarations

Conflict of interest Peggy Gatignol is co-author of the MBLF tool. As such, she participated in the development of the tool and its standardization. Dr. Arnulf had no link of interest related to the study. Outside the study, she took part in an advisory board of IDORSIA in 2020. Dr. Attali had had no link of interest related to the study. Outside the study, she took part in an advisory board of BIOPROJET in 2021. Dr. Similowski reports personal fees for consulting and teaching activities from ADEP Assistance, AstraZeneca France, Chiesi France, KPL consulting, Lungpacer Inc., OSO-AI, TEVA France, Vitalaire, all outside the submitted work. The others authors declare no conflict of interest with respect to the research.

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