

ORIGINAL RESEARCH

Orofacial dysfunction screening examinations in children with sleep-disordered breathing symptoms

Dao Anh Hoang^{1,3}, Van Nhat Thang Le¹, Tam Minh Nguyen², Triin Jagomägi^{3,*}

¹Faculty of Odonto-Stomatology, Hue University of Medicine and Pharmacy, Hue University, 49120 Hue, Vietnam

²Department of Family Medicine, Hue University of Medicine and Pharmacy, Hue University, 49120 Hue, Vietnam

³Institute of Dentistry, Faculty of Medicine, University of Tartu, 50090 Tartu, Estonia

***Correspondence**

triin.jagomagi@ut.ee
(Triin Jagomägi)

Abstract

Orofacial myofunctional disorders (OMD) and sleep-disordered breathing (SDB) may present as comorbidities. Orofacial characteristics might serve as a clinical marker of SDB, allowing early identification and management of OMD and improving treatment outcomes for sleep disorders. The study aims to characterize OMD in children with SDB symptoms and to investigate possible relationships between the presence of various components of OMD and symptoms of SDB. A cross-sectional study of healthy children aged 6–8 from primary schools was conducted in central Vietnam in 2019. SDB symptoms were collected using the parental Pediatric Sleep Questionnaire, Snoring Severity Scale, Epworth Daytime Sleepiness Scale, and lip-taping nasal breathing assessment. Orofacial myofunctional evaluation included assessment of tongue mobility, as well as of lip and tongue strength using the Iowa Oral Performance Instrument, and of orofacial characteristics by the protocol of Orofacial Myofunctional Evaluation with Scores. Statistical analysis was used to investigate the relationship between OMD components and SDB symptoms. 487 healthy children were evaluated, of whom 46.2% were female. There were 7.6% of children at high risk of SDB. Children with habitual snoring (10.3%) had an increased incidence of restricted tongue mobility and decreased lip and tongue strength. Abnormal breathing patterns (22.4%) demonstrated lower posterior tongue mobility and lower muscle strength. Daytime sleepiness symptoms were associated with changes in muscle strength, facial appearance, and impaired orofacial function. Lower strengths of lip and tongue or improper nasal breathing were more likely to be present in children with reported sleep apnea (6.6%). Neurobehavioral symptoms of inattention and hyperactivity were linked to anomalous appearance/posture, increases in tongue mobility and oral strength. This study demonstrates a prevalence of orofacial myofunctional anomalies in children exhibiting SDB symptoms. Children with prominent SDB symptoms should be considered as candidates for further orofacial myofunctional assessment.

Keywords

Orofacial dysfunction; Sleep-disordered breathing; Screening examination

1. Introduction

Sleep is essential for growth, development, learning, and well-being in children and adolescents. Sleep problems may be caused, among other things, by sleep-disordered breathing (SDB) which encompasses a spectrum of conditions characterized by abnormal breathing: from snoring to upper airway resistance syndrome up to obstructive sleep apnea (OSA) [1]. SDB in children has been associated with various symptoms [2–9]. Common symptoms reported by patients include morning tension-type headaches, excessive daytime fatigue, restless sleep, heavy snoring, poor concentration, mood disturbance, aggressiveness and attention-deficit/hyperactivity disorder.

According to the European Respiratory Society, pediatric OSA is defined as a syndrome of upper airway dysfunction

during sleep, characterized by snoring or increased respiratory effort resulting from increased upper airway resistance and pharyngeal collapsibility [10]. Reported prevalence of OSA in children and adolescents ranges between 1% and 5% but may be underdiagnosed [11–13]. Previous studies indicate that children with OSA tend to exhibit mouth breathing and a lower tongue position which causes an imbalance of muscular forces of the cheek and tongue [14]. Tongue position has also been shown to correlate with the severity of OSA [15]. These studies demonstrate that pediatric patients with OSA tend to present a degree of orofacial dysfunction—although the orofacial myofunctional status of such patients has not been fully evaluated.

Orofacial dysfunction is also known as orofacial myofunctional disorders (OMD)—disorders of the oral and facial mus-

culature (lips, jaw, tongue and oropharynx) [16, 17]. OMD lead to anomalies of appearance, posture and mobility of the lips, tongue, mandible, and cheeks [18]. Several studies that have investigated the association between SDB and OMD report a significant relationship between the two [19, 20]. However, none of the studies investigated this relationship in the context of primary screening. Until recently, screening examinations in children with SDB symptoms did not focus on the possible presence of OMD. Yet, early identification of OMD in patients with SDB symptoms may be useful for selecting an effective intervention.

To the best of the authors' knowledge, there has been no study that evaluated the relationship between OMD and SDB symptoms in children who underwent clinical screening examinations. The research question of this paper is whether OMD and SDB are likely to present as comorbidities. As early identification of SDB-related anomalies promises better treatment outcomes, the study at hand investigated the presence of orofacial dysfunction by administering screening examinations to children with SDB symptoms.

2. Materials and methods

2.1 Study population and design

In this study, the multi-stage stratified random sampling method was used to select a sample to cover urban as well as rural regions. Two primary schools were randomly selected from the list of primary schools in Thua Thien Hue province (one in the rural area and one in Hue city). The inclusion criteria were enrolment in primary school and being aged 6–8 years. Exclusion criteria included a history of frenectomy, myofunctional therapy, oral motor disabilities (dysphagia, facial or lingual nerve paralysis), tongue, lip, or cheek surgery, and any prior apnea treatment. The data were collected from February 2019 to May 2019. In total, 487 pupils were eligible to participate.

2.2 Data collection

2.2.1 Assessment of SDB symptoms

Data including age, height, weight, medical history, neck circumference and hand strength were collected from the participants. Information on symptoms and signs related to SDB was obtained from parents/caregivers and children by using a combination of subjective questionnaires (on sleep and daytime behavior, snoring severity, and daytime sleepiness) [21–25], a visual clinical evaluation of upper airway obstructions (Mallampati classification and Friedman tongue position, tonsil size according to Brodsky scale) [26] and an objective test (nasal breathing assessment) [27].

The parents/caregivers completed the non-validated Vietnamese version (created using the forward-backward translation method recommended by the World Health Organization) of the 22-item Sleep-Related Breathing Disorder scale of the Pediatric Sleep Questionnaire (PSQ) on behalf of their children [21, 22]. The symptom items—each previously shown by polysomnography to correlate with confirmed pediatric OSA—included snoring frequency, loud snoring, observed apnea, breathing difficulty during sleep, daytime sleepiness,

inattentive or hyperactive behavior and other OSA features. Response options were “yes” = 1, “no” = 0, and “don't know” = missing. The mean response on non-missing items provided a score in the range of 0 to 1. Eight or more affirmative responses (cut-off value of 0.33) were considered abnormal and indicative of an obstructive sleep-related breathing disorder [21]. In addition, the participants were asked to fill in the Epworth Sleepiness Scale (ESS) questionnaire concerning their propensity to fall asleep in eight specific situations (on a scale of 0 to 3). The ESS score indicates the degree to which children complained of sleepiness. A score of more than 10 was considered a marker of possibly excessive daytime sleepiness [23]. The third questionnaire—the Severity Snoring Scale (SSS)—was filled in by the parents/caregivers (and supplemented by children where necessary) who reported on the loudness, frequency and periodicity of the snoring sound [24]. A total SSS score that equalled or exceeded 7 was considered to indicate a high risk of OSA [25].

To evaluate upper airway obstructions, tonsil size assessments were conducted by visually inspecting the tonsils and recording a corresponding score using the Brodsky grading scale (1 to 4) depending on the percentage of the oropharyngeal airway occupied by the tonsils. Cases of greater than 50% obstruction (grade 3 and grade 4) were classified as palatine tonsil hypertrophy [28]. Furthermore, both Mallampati and Friedman tongue position scales were used to assess tongue position relative to other soft tissues in the back of the throat [29]. For nasal breathing assessment, each participant underwent the lip taping test. Participants were instructed to seal their lips and mouth with tape and maintain nasal breathing for three minutes [27]. The test assesses the patient's capacity for comfortable nasal breathing and the possible presence of a nasal obstruction or mouth breathing habit. Subjects were considered to have passed or to be unable to complete the test according to whether or not they could successfully complete the test.

2.2.2 Evaluation for orofacial myofunctional disorders

With respect to orofacial dysfunction, three parameters were measured: tongue mobility, maximal lip and tongue strength using the Iowa Oral Performance Instrument (IOPI) and orofacial characteristics determined by means of a validated protocol of the Orofacial Myofunctional Evaluation with Scores (OMES) [17].

To assess tongue mobility, tongue range of motion ratio (TRMR) was calculated based on measurements of the functional movements of tongue-tip-to-incisive-papillae (TIP) and lingual-palatal suction (LPS) [30, 31]. All measurements were obtained using a tongue range of motion instrument (Great Lakes Orthodontics; MI, USA) with the subjects sitting upright in a natural position with a horizontal visual axis. First, the interincisal distance at maximum mouth opening (CMO) without pain or discomfort was measured. Next, the interincisal distance at maximum mouth opening with TIP and at mouth opening while maintaining contact between the tongue body and the palate in LPS was obtained. Finally, tongue range of motion deficit (TRMD) was calculated as, respectively, the difference between CMO and TIP or between

CMO and LPS [32]. TRMR—the ratio of TIP or LPS to CMO—reflects the mobility of the anterior or posterior part of the tongue.

For maximal lip and tongue strength, objective measurements were conducted using the IOPI (Medical LLC, Carnation, WA, USA). The study followed the protocol described by Potter and Short [33]. Measurements were taken with participants sitting in an upright position, three times for 5 seconds with a 30-second break between each measurement. The highest of the three measurements was recorded as the participant's lip or tongue strength. All strength measurements were expressed in kilopascals (kPa). Tongue strength represents the muscle tone of the genioglossus muscle while lip strength represents that of the buccinator muscle. Children were asked to raise their tongue and squeeze the instrument's bulb against the palate as hard as they could for approximately 3 seconds. Anterior tongue strength was obtained by measuring maximum tongue elevation pressure with the bulb positioned just behind the alveolar ridge while the bulb stem was held by the examiner immediately before the central incisors. Posterior tongue strength was obtained by measuring maximum tongue elevation pressure with the bulb positioned on the midline of the tongue at the sulcus terminalis. The highest value of anterior and posterior strength was noted as the respective maximum strength measured during the exercise. For lip strength measurement, participants were asked to squeeze the IOPI bulb against the buccal surface of the teeth by pursing the lips as hard as possible when the bulb was located between the cheek and closed teeth.

For orofacial myofunctional status evaluation, children were individually examined according to the OMES protocol [17]. Total OMES scores were calculated across three categories: (1) posture (including facial symmetry, cheeks, mandible, lips, tongue, and hard palate); (2) mobility (assessed by having the subjects perform 4–6 movements with each component of the lips, tongue, cheeks or mandible) and (3) functions (including breathing mode, deglutition and mastication). Total OMES scores have a range from 32 to 104, with higher values indicating better orofacial myofunctional condition and a normal stomatognathic system. As the hard palate score was not included in the analysis in this study, the maximum possible OMES score was 101 instead of 104.

2.3 Data reproducibility and reliability

To determine errors in tongue function measurements (*i.e.*, tongue mobility and tongue strength), repeated measurements were taken on 25 random children by the same investigator after at least one month. The random error was calculated using Dahlberg's equation [34]. A systematic error was assessed using the paired *t*-test, for $p < 0.01$. As a result, the systematic measurement error did not exceed 2 mm for tongue mobility or 1 kPa for tongue strength. The correlation was higher than 0.90 for all measures. The test-retest random error was much lower than the intra-examiner standard deviation, showing the good reproducibility of measures. There were no statistically significant differences between the two measurements. Therefore, the systematic measurement errors were considered to be insignificant.

To eliminate interexaminer variation, each researcher was responsible for evaluating variables simultaneously and independently across all samples. Tongue function and orofacial myofunction evaluation were measured by one trained dentist (main investigator), while airway obstruction assessment (*i.e.*, tonsil size and breathing mode) was independently performed by a trained orthodontist.

Measurement reliability was assessed during the study on occasions that involved taking repeated measurements on 25 randomly selected subjects. The intra-class correlation coefficients (ICC) in tongue function measurements were as high across the entire sample as greater than 0.80. Clinical diagnostic measures of orofacial dysfunction were determined for a second time with the following ICC: airway obstruction assessment: tonsil size = 0.90, breathing mode = 0.80; OMES categories: appearance/posture = 0.90, mobility = 0.80, functions = 0.85.

For the pediatric sleep questionnaire, test-retested reliability data were collected on a separate sample of 25 children whose parents completed the questionnaire items twice, first on the day of their children's initial evaluation and then again by email approximately one month later. Cronbach's alpha for each item was: snoring scale, 0.86; sleepiness scale, 0.65; behavior scale, 0.84; and SDB scale, 0.89.

2.4 Statistical analysis

Statistical analyses were performed using IBM SPSS Statistics (version 22; IBM Corp., Armonk, NY, USA). Continuous variables were summarized as mean \pm standard deviation (SD) for normally distributed continuous variables, or median and interquartile range (25th–75th percentile) for non-normally distributed continuous variables. Categorical variables were summarized as frequencies and percentage values. For normally distributed data, univariate analysis was performed by Pearson's chi-square and independent *t*-test to assess correlations between nominal or continuous covariates of orofacial symptoms characteristic of SDB; for non-normal distributions, the Fisher exact test or Mann-Whitney test was used. Statistical significance was set at a $p < 0.05$.

3. Results

As presented in Table 1, the study included 225 female and 262 male children (46.2% and 53.8% respectively) with a mean age of 7.5 ± 0.5 years. The participants' mean BMI percentile was 41.1 ± 37.1 , the number of obese participants was 60 (12.3%). Enlarged tonsils were observed in 17.3% of the children. A high tongue position (according to Friedman) was subjectively assessed to be present in 47.3% of participants.

Table 2 depicts a wide range of SDB symptoms which was reported by parents, which suggested that 7.6% of children were at high risk of SDB (8 or more "yes" responses on the PSQ). Habitual snoring (usually snoring and always snoring during sleep) was reported for 10.3% of the children and moderate to severe snoring for another 2.5%. Breathing problems—*i.e.*, trouble breathing during sleep—were reported in 8%, and observed stops in breathing suggesting apnea (none of the subjects had previously been diagnosed with apnea)

TABLE 1. Baseline characteristics of participants with SDB symptoms.

Baseline characteristics	<i>N</i> = 487
Sex, male (%)	262 (53.8)
Age, mean (SD)	7.5 (0.5)
Neck circumference (mm), mean (SD)	27.2 (2.3)
BMI percentile, mean (SD)	41.1 (37.1)
Obesity* (%)	60 (12.3)
Hand strength (kPa), mean (SD)	10.0 (2.6)
High tongue position (Grade III and IV Friedman) (%)	224 (47.3)
High tongue position (Grade III and IV Mallampati) (%)	220 (48.2)
Tonsil enlargement (%)	109 (22.9)

BMI: Body Mass Index; SD: standard deviation.

*Obesity means BMI \geq 95th percentile.

in 6.6%. The prevalence of mouth breathing during wakefulness was 22.4%; 9% of the children found it difficult to breathe through the nose during the lip seal test. Daytime sleepiness was common with 38.6% of children who reported feeling unrefreshed in the morning; 28.1% were reported to be hard to wake. Behavioral problems ranged from being easily distracted (29.8%) and fidgety (26.7%) to agitated (9.9%). Developmental symptoms (occasional bedwetting) had been observed in 11.3% of the children and 7.6% had experienced delayed growth since birth.

Table 3 shows the principal anomalies of orofacial myofunctional characteristics in habitually snoring children. Tongue strength was significantly lower in snorers ($p < 0.05$). As for tongue mobility in the elevating movement of the anterior part of the tongue, mouth opening reduction was significantly higher in snorers whose TRMR was significantly lower ($p < 0.05$). Snoring intensity tended to correlate significantly with reduced degrees of anterior tongue mobility and increased limitation of movement of the anterior part of the tongue.

A significant association between orofacial myofunctional characteristics and breathing-related symptoms of SDB is shown in Table 4. Only posterior tongue mobility or mobility of the tongue base showed a significant divergence (reduced mouth opening with tongue in LPS and a lower TRMR) in children with dry mouth on awakening and in children with daytime mouth breathing, the latter displaying higher degrees of functional reduction. Lip and tongue strength were significantly lower in children who experienced apnea during sleeping ($p < 0.05$). The breathing subscores of the OMES protocol were lower in children with stops in sleep-time breathing ($p < 0.001$) or mouth breathing symptoms ($p < 0.05$). Children who were unable to complete the 3-minute lip-sealing test, implying difficulty in nasal breathing, had lower breathing subscores ($p < 0.001$).

Individuals with daytime sleepiness symptoms showed a reduction in muscle tone (reduced lip strength in the “problems with sleepiness” and reduced tongue strength in the “being hard

to wake” category, respectively) ($p < 0.05$). In the assessment of orofacial function, significantly lower posture subscores were recorded for children who were reported to sleepy and wet the bed while significantly lower breathing subscores were reported for those with a history of delayed growth ($p < 0.001$). Children with excessive daytime sleepiness had lower total OMES scores ($p < 0.05$) (Table 5).

Table 6 shows that children with behavioral symptoms of SDB (fidgety or interrupting others) displayed a significant increase in tongue mobility. Lip muscle strength was significantly higher in children who were fidgety and posterior tongue strength in those who tended to appear agitated or not to listen to others. In orofacial function evaluation, reduced subscores were recorded for—children who appeared not to listen to others (in comparison with the posture subscores), and who tended to interrupt others (in comparison with the posture and function subscores).

4. Discussion

Recent studies suggest that applying orofacial myofunctional therapy as a treatment of OMD also relieves OSA in children [35] and adults [36]. It is thus important for the clinician to be able to identify tendencies in the orofacial aspects of primary school children that indicate a risk of sleep impairment. The study at hand assessed primary school children in terms of the child’s symptoms of SDB as reported by the parents/caregivers and the child’s orofacial myofunctional status (appearance/posture, mobility, and functions), mobility of anterior or posterior part of the tongue and lip and tongue strength. A wide variety of SDB symptoms was reported by the participants’ parents, showing 7.6% of healthy children to be at high risk of SDB. All symptoms associated with OSA were also present in healthy children with occurrence rates varying from very low at 6.4% (for “sleepy as reported by a teacher”) to very frequent at 42.5% (for “dry mouth on awakening”). However, few prominent symptoms of SDB such as habitual snoring (14.3%), exhibiting stops in breathing during sleep (6.6%), showing signs of mouth breathing (22.4%–42.5%) or of daytime sleepiness (6.4%–28.1%) or developmental concerns (11.3% for occasional bed-wetting and 7.6% for having a history of delayed growth) displayed a significant relationship to orofacial dysfunction and reported behavioral problems (not listening, being fidgety, showing agitation—at 12.5%, 26.7%, and 9.9% respectively). Recent evidence has emphasized that even mild forms of SDB could have important repercussions in children, who are particularly vulnerable to harmful effects of disturbed sleep which may lead to impairment of growth or have an adverse impact on the development of behavioral and cognitive abilities [37]. The fact that our study, which focused on healthy children, revealed a widespread presence of signs and features of SDB suggests that these may frequently not be appropriately recognized or diagnosed in a large part of the young population. This might be due to either difficulties of access to diagnostic methods or weak perception of sleep problems [38].

With respect to neuromuscular factors of pediatric OSA the finding of this study is that the relationship between SDB and the role of the stomatognathic system should be inves-

TABLE 2. SDB symptoms as described in the questionnaire administered to the participants.

Category	Features	N (%)
Snoring frequency		
	Usually snores	29 (6.0)
	Always snores	21 (4.3)
Snoring quality		
	Snores loudly	37 (7.6)
	Heavy or loud breathing	43 (8.8)
Moderate to severe snoring		
	SSS score over seven	12 (2.5)
Breathing problems		
	Trouble breathing during sleep	39 (8.0)
	Stops breathing during sleep	32 (6.6)
Mouth breathing		
	Daytime mouth breathing	109 (22.4)
	Dry mouth on awakening	207 (42.5)
Nasal breathing difficulty		
	Unable to complete the lip seal test	44 (9.0)
Daytime sleepiness		
	Feeling unrefreshed in the morning	188 (38.6)
	Problem with somnolence	68 (14.0)
	Sleepy as reported by a teacher	31 (6.4)
	Difficult to wake in the morning	137 (28.1)
Inattention/hyperactivity		
	Does not seem to listen when spoken to	61 (12.5)
	Difficulty organizing tasks and activities	54 (11.1)
	Easily distracted by external stimuli	145 (29.8)
	Fidgets with hands or feet	130 (26.7)
	Agitated (seems restless and cannot remain still when seated)	48 (9.9)
	Interrupts or intrudes on others	102 (20.9)
Other symptoms		
	Occasionally wets the bed	55 (11.3)
	Morning headache	73 (15.0)
	Delayed growth since birth	37 (7.6)
	Overweight	75 (15.4)
Excessive daytime sleepiness		
	ESS score over ten	19 (3.9)
Number of children at high-risk of SDB		
	Eight or more "yes" responses	37 (7.6)

ESS: Epworth Sleepiness Scale; SDB: sleep-disordered breathing.

TABLE 3. Orofacial myofunctional characteristics by snoring symptoms.

Category	N	Anterior tongue mobility			Posterior tongue mobility		Tongue strength, kPa		
		TIP, mm	TRMD, mm	TRMR, %	TRMD, mm	TRMR, %	Anterior strength	Posterior strength	Maximum strength
Usually snores									
No	458	26.9 ± 5.0	18.8 ± 6.0	59.2 ± 11.4	29.4 ± 6.1	35.9 ± 10.7	45.5 ± 11.5	41.3 ± 12.7	46.4 ± 11.5
Yes	29	25.5 ± 6.6	21.2 ± 6.6	54.6 ± 13.8	31.5 ± 5.0	32.6 ± 8.6	40.4 ± 11.3	35.8 ± 12.1	41.4 ± 11.2
<i>p</i> -value*		ns	0.038	0.041	ns	ns	0.022	0.024	0.024
Moderate to severe snoring									
No	475	26.9 ± 5.0	18.9 ± 6.0	59.1 ± 11.5	29.5 ± 6.1	35.8 ± 10.7	45.2 ± 11.6	41.0 ± 12.8	46.1 ± 11.6
Yes	12	23.8 ± 6.8	22.0 ± 6.9	52.0 ± 14.3	30.8 ± 4.7	32.8 ± 7.9	42.8 ± 9.9	37.9 ± 10.8	43.4 ± 10.2
<i>p</i> -value*		0.036	ns	0.036	ns	ns	ns	ns	ns

Significance was considered at the *p*-value < 0.05.

Abbreviations: ns: non-significant; TIP: tongue-tip-to-incisive-papillae; TRMD: tongue range of motion deficit; TRMR: tongue range of motion ratio. *Mann-Whitney U test.

TABLE 4. Orofacial myofunctional characteristics by breathing-related symptoms.

Category	N	Posterior tongue mobility			Lip strength	Tongue strength, kPa			Function (OMES protocol)
		LPS, mm	TRMD, mm	TRMR, %	kPa	Anterior strength	Posterior strength	Maximum strength	Breathing score
Stop breathing									
No	455	16.3 ± 4.7	29.4 ± 6.1	29.4 ± 6.1	19.9 ± 4.0	45.6 ± 11.4	41.4 ± 12.7	46.4 ± 11.4	3.0 ± 0
Yes	32	15.4 ± 3.7	30.9 ± 5.6	30.9 ± 5.6	18.1 ± 4.4	39.3 ± 12.4	35.2 ± 12.4	40.8 ± 12.1	2.9 ± 0.2
<i>p</i> -value*		ns	ns	ns	0.014	0.003	0.008	0.008	<0.001
Daytime mouth breathing									
No	378	16.4 ± 4.8	28.2 ± 6.3	36.2 ± 11.1	19.8 ± 3.9	45.8 ± 11.2	41.3 ± 12.3	46.4 ± 11.3	3.0 ± 0.1
Yes	109	15.6 ± 4.0	30.4 ± 5.2	34.1 ± 8.6	19.8 ± 4.6	43.1 ± 12.5	39.8 ± 14.3	44.8 ± 12.5	2.9 ± 0.2
<i>p</i> -value*		ns	ns	0.033	ns	ns	ns	ns	0.003
Dry mouth on awakening									
No	280	16.6 ± 4.7	28.8 ± 6.0	36.8 ± 10.7	19.8 ± 4.0	45.2 ± 11.3	40.9 ± 12.3	45.8 ± 11.4	2.9 ± 0.2
Yes	207	15.7 ± 4.5	30.4 ± 6.1	34.4 ± 10.4	19.8 ± 4.1	45.1 ± 11.9	41.0 ± 13.4	46.4 ± 11.8	3.0 ± 0.2
<i>p</i> -value*		0.043	0.004	0.014	ns	ns	ns	ns	ns
Nasal breathing difficulty									
No	436	16.2 ± 4.7	35.8 ± 10.7	29.5 ± 6.0	19.8 ± 4.1	45.2 ± 11.7	40.8 ± 12.8	46.1 ± 11.6	3.0 ± 0
Yes	40	16.1 ± 4.3	35.6 ± 10.5	29.7 ± 6.5	19.7 ± 4.4	44.6 ± 10.5	42.3 ± 13.0	45.5 ± 11.0	2.9 ± 0.2
<i>p</i> -value*		ns	ns	ns	ns	ns	ns	ns	<0.001

Significance was considered at the *p*-value < 0.05.

Abbreviation: LPS: Maximum mouth opening with the tongue in LPS; TRMD: tongue range of motion deficit; TRMR: tongue range of motion ratio; OMES: Orofacial Myofunctional Evaluation Protocol with Scores; ns: non-significant. *Mann-Whitney U test.

TABLE 5. Orofacial myofunctional characteristics by daytime sleepiness and developmental symptoms.

Category	N	Lip strength, kPa		Tongue strength, kPa			Orofacial function (OMES protocol)		
				Anterior	Posterior	Maximum	Posture	Breathing	OMES score
Problem with sleepiness									
No	419	19.9 ± 4.0	45.4 ± 11.4	41.3 ± 12.6	46.3 ± 11.5	14.9 ± 0.3	2.9 ± 0.3	99.4 ± 2.8	
Yes	68	18.8 ± 4.3	43.4 ± 12.2	38.5 ± 13.6	44.4 ± 12.1	15.0 ± 0.2	2.9 ± 0.2	99.5 ± 1.4	
<i>p</i> -value*		0.029	ns	ns	ns	ns	ns	ns	
Hard to wake up									
No	350	20.0 ± 3.8	45.9 ± 11.1	41.8 ± 12.2	46.8 ± 11.0	14.9 ± 0.3	2.9 ± 0.2	99.5 ± 2.7	
Yes	137	19.2 ± 4.7	43.3 ± 12.4	38.8 ± 14.0	44.2 ± 12.7	14.9 ± 0.3	2.9 ± 0.2	99.3 ± 2.9	
<i>p</i> -value*		ns	0.026	0.029	0.034	ns	ns	ns	
Reported sleepy									
No	446	19.8 ± 4.1	45.2 ± 11.4	41.0 ± 12.6	46.1 ± 11.4	15.0 ± 0	2.9 ± 0.2	99.8 ± 1.2	
Yes	30	18.9 ± 3.8	44.4 ± 13.5	40.0 ± 15.7	45.2 ± 13.8	14.9 ± 0.3	2.9 ± 0.2	99.4 ± 2.8	
<i>p</i> -value*		ns	ns	ns	ns	<0.001	ns	ns	
Occasionally wets the bed									
No	421	19.8 ± 4.1	45.3 ± 11.4	41.0 ± 12.6	46.2 ± 11.4	15.0 ± 0	2.9 ± 0.3	99.7 ± 1.2	
Yes	55	19.6 ± 3.8	44.1 ± 13.5	40.3 ± 15.7	45.0 ± 13.8	14.9 ± 0.3	2.9 ± 0.2	99.4 ± 2.9	
<i>p</i> -value*		ns	ns	ns	ns	<0.001	ns	ns	
Delayed growth									
No	442	19.8 ± 4.1	45.2 ± 11.6	40.9 ± 12.8	46.1 ± 11.7	14.9 ± 0.3	3.0 ± 0	99.7 ± 1.2	
Yes	34	19.7 ± 4.3	44.6 ± 10.5	41.8 ± 12.0	45.8 ± 10.28	14.9 ± 0.3	2.9 ± 0.2	99.4 ± 2.8	
<i>p</i> -value*		ns	ns	ns	ns	ns	<0.001	ns	
Excessive daytime sleepiness									
No	468	19.8 ± 4.1	45.1 ± 11.5	40.8 ± 12.8	46.0 ± 11.6	14.9 ± 0.2	2.9 ± 0.2	100.1 ± 1.1	
Yes	19	18.4 ± 4.4	45.3 ± 12.5	43.2 ± 12.3	46.7 ± 11.1	14.9 ± 0.3	2.9 ± 0.2	99.4 ± 2.8	
<i>p</i> -value*		ns	ns	ns	ns	ns	ns	0.021	

Significance was considered at the *p*-value < 0.05. *Mann-Whitney U test. OMES: Orofacial Myofunctional Evaluation Protocol with Scores; ns: non-significant.

TABLE 6. Orofacial myofunctional characteristics by behavioral symptoms.

Category	N	Anterior tongue mobility		Lip strength, kPa	Posterior tongue strength, kPa	Orofacial function (OMES protocol)			
		TRMD, mm	TRMR, %			Posture	Mastication	Function score	
Does not listen									
No	426	19.0 ± 6.0	58.8 ± 11.6	19.8 ± 4.0	40.5 ± 12.8	14.9 ± 0.3	15.8 ± 0.9	27.7 ± 1.4	
Yes	61	18.6 ± 6.2	59.4 ± 11.9	19.9 ± 4.7	44.4 ± 12.3	14.9 ± 0.1	15.8 ± 0.4	27.8 ± 1.2	
<i>p</i> -value*		ns	ns	ns	0.027	0.004	ns	ns	
Agitated									
No	439	18.9 ± 6.1	58.9 ± 11.7	19.8 ± 3.9	40.5 ± 12.7	14.9 ± 0.3	15.7 ± 0.9	27.8 ± 1.4	
Yes	48	19.2 ± 5.8	58.2 ± 10.4	19.8 ± 5.3	45.1 ± 13.2	14.9 ± 0.3	15.8 ± 0.5	27.6 ± 1.3	
<i>p</i> -value*		ns	ns	ns	0.019	ns	ns	ns	
Fidgets									
No	357	19.4 ± 6.2	58.1 ± 12.0	19.5 ± 3.8	40.3 ± 12.5	14.9 ± 0.2	15.7 ± 0.8	27.8 ± 1.7	
Yes	130	17.8 ± 5.5	61.0 ± 10.2	20.5 ± 4.7	42.7 ± 13.5	14.9 ± 0.3	15.9 ± 0.7	27.6 ± 1.2	
<i>p</i> -value*		0.011	0.016	0.027	ns	ns	0.049	ns	
Interrupts									
No	385	19.3 ± 6.2	58.3 ± 11.8	19.8 ± 4.0	41.0 ± 12.8	14.9 ± 0.3	15.7 ± 0.8	27.8 ± 1.7	
Yes	102	17.5 ± 5.4	61.3 ± 10.6	19.9 ± 4.5	40.8 ± 12.6	14.9 ± 0.2	15.9 ± 0.7	27.5 ± 1.2	
<i>p</i> -value*		0.008	0.018	ns	ns	0.02	ns	0.049	

Significance was considered at the *p*-value < 0.05. *Mann-Whitney U test. TRMD: tongue range of motion deficit; TRMR: tongue range of motion ratio; OMES: Orofacial Myofunctional Evaluation Protocol with Scores; ns: non-significant.

tigated in terms of tongue mobility and muscular strength. The study demonstrated that children affected by sleep problems (habitual snoring, mouth breathing) tend to present a reduction in tongue mobility and a reduced tongue range of motion, especially in the upward direction. Recent literature suggests that the short lingual frenulum, an anatomical condition characterized by anomalies that result in various degrees of restricted tongue mobility, is a risk factor for SDB in school-age children [39, 40]. By not assessing tongue mobility [41] or its limitations expressed as TRMD, previous studies that reported short lingual frenulum (defined by less than 16 mm of Kotlow's free tongue length [42]) exposed themselves to a significant risk of measurement errors. The tongue needs to have full range and accuracy of movement in accordance with the demands of its tasks in order not to induce pharyngeal collapse or upper airway obstruction due to either its increased volume, upright position or imbalanced interaction with surrounding tissues [43]. Interestingly, we found that children with concerns of hyperactivity (fidgets) and behavioral performance (habitually interrupting others during conversation) had a significant increase in anterior tongue mobility in the upward direction. This is consistent with the result of a previous retrospective study which showed that children with hypermobility spectrum disorders have prominent co-occurring symptoms of attention-deficit/hyperactivity disorder [44] although the etiology of the phenomenon remains unclear.

The study at hand shows children with SDB symptoms to have reduced orofacial muscle strength as measured by the IOPI. This observation is similar to the results of a previous study which assessed the role of orofacial myofunctional therapy [45]. In our study, the reduction of muscle tone varied depending on the category of symptoms and on the type of muscle. While snorers tended to have reduced tongue strength, subjects with signs of daytime sleepiness ("problems with sleepiness" or "hard to wake up") showed reductions in lip as well as tongue strength. In particular, children with reported sleep apnea displayed lower orofacial force in both the buccinator and the anterior or posterior genioglossus. This may contribute to airway collapse susceptibility at the tongue base, which is one of the underlying pathologies in OSA [46]. Such tongue base collapses are due to improper tongue position and volume or to a reduction of muscle tone during sleep [47, 48]. However, in our study, children having behavioral symptoms (inattention and hyperactivity) showed significantly stronger force in the lip and tongue than those without such symptoms. Similar to tongue mobility, the etiology of phenomena remains unclear. In a recent report, Birk *et al.* [46] concluded that maximum isometric tongue force in patients with OSA does not seem to differ from that observed in healthy subjects (although patients with OSA displayed stronger isometric tongue force, the figures were not significant). Yet, in that study, tongue force was measured differently: in an anterior/posterior direction in a protrusion task instead of the tongue elevation and lip compression tasks of our study. Thus, further studies that test all directions of tongue force and all tongue tasks should be performed in order to verify the presence of divergence (either lower or higher maximum isometric orofacial force) in patients with OSA.

During orofacial myofunctional assessment by the OMES

protocol, children with specific SDB symptoms mainly presented variations in appearance/posture and breathing function. Most of the variations observed were consistent with data from mouth-breathing children who had significantly lower mean breathing scores than those without mouth breathing [20]. Daytime mouth breathing (characterized by supplementary mouth breathing) is an early sign of SDB [49]. For children classified as mouth breathers, the lip seal test was an important criterion to diagnose whether mouth breathing was by habit or obstruction [28]. In our study, mouth breathers by obstruction who could not complete the 3-minute lip seal test were likely to exhibit anomalies in their breathing mode. The presence of habitual mouth breathing (with difficulties in nasal breathing) associated with an improper breathing pattern (as shown by the low breathing component scores described in the study at hand) emphasizes the importance of clinical assessment of route of breathing in children with symptoms associated with OSA. This study found statistically significant associations between appearance/posture subscores and reported sleepiness, bed wetting, and behavioral parameters (not listening to others or interrupting them). The OMES assigns scores to changes in soft structures such as cheeks, lips, facial symmetry and the tongue, as well as to hard structures (the jaw and palate) [17]. Children with SDB symptoms (reported sleepiness, bed wetting, behavioral divergences) presented a significantly lower score of this component, indicating anomalies in appearance/posture (either lip incompetence, open mouth posture, tongue protrusion or posture between dental arches, signs of flaccid/drooping cheeks or facial asymmetry). Structural etiologies such as low tongue resting posture and lips-apart open mouth posture may also physically manifest as mouth breathing [45], and may increase the risk of snoring and OSA in children. A study assessing the diagnostic value of cheek appearance for determining the presence of sleep apnea found that cheek appearance served as a predictor of the risk of moderate to severe OSA [50]. As our study did not analyze the SDB symptoms based on specific OMES items, it is not possible to identify the item or items responsible for a lower overall score. However, the correlation of lower posture subscores and SDB symptoms appears to indicate divergences of appearance and posture in children with some manifestations of SDB. The diagnostic recognition of facial features which could be pathognomonic of OSA has been mentioned rarely in reviewed literature [50]. Learning to recognize elements of appearance/posture that suggest an OMD associated with SDB symptoms could prove an important addition to the clinician's repertory of diagnostic tools.

The study at hand has several limitations. First, it was designed as a cross-sectional observation—which necessitates a further cohort study to confirm the association between OMD and SDB in screening examinations. Second, its results are susceptible to reporter bias due to parental observations of sleep and behavior as well as to concerns related to recognition of SDB symptoms and orofacial dysfunction. The study mainly relied on parents' and children's reports rather than objectively collected sleep measures such as polysomnography. Because access to polysomnography is limited, PSQ was used as a valid and reliable instrument for identifying the

risk of SDB. In addition to PSQ, the presence of symptoms of SDB was evaluated in parallel by an objective test (the lip seal test) to predict difficulties with nasal breathing. Furthermore, we adopted a validated OMES protocol to minimize the bias related to clinical OMD evaluation. Reproducible and reliable measure of muscle strength (IOPI) and a reliable tool for functional assessment of tongue mobility (TRMR) were used. Finally, the etiology of increases tongue mobility and oral muscle tone in children with SDB symptoms (including inattention and hyperactivity) remain unclear. Further studies should be performed to clarify these issues.

5. Conclusions

The study's screening examination revealed a statistically significant presence of orofacial myofunctional anomalies in children with SDB symptoms. Children with habitual snoring are more likely to suffer from restrictions of tongue mobility as well as reduced lip and tongue strength. Abnormal breathing patterns had lower posterior tongue mobility and lower muscle strength. Daytime sleepiness symptoms were associated with changes in oral muscle strength, facial appearance, and impaired orofacial function. Lower strengths of lip and tongue or improper nasal breathing were more likely to be present in children with reported sleep apnea. Neurobehavioral symptoms of inattention and hyperactivity were linked to anomalies in appearance/posture, as well as an increase in tongue mobility and oral muscle tone. The results of this study suggest that children with prominent SDB symptoms should be considered as candidates for further orofacial myofunctional assessment.

AVAILABILITY OF DATA AND MATERIALS

The data presented in this study are available on reasonable request from the corresponding author.

AUTHOR CONTRIBUTIONS

TJ, TMN and DAH—designed the research study. DAH and TMN—performed the research. DAH and TMN—analyzed the data. DAH, VNTL and TJ—wrote the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

Written informed consent was obtained from all children and parents/caregivers before their enrolment in the study. The study was conducted in accordance with the Declaration of Helsinki, and the protocol was approved by the Institutional Review Board of Hue University of Medicine and Pharmacy (No. H2018/16).

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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