Accepted Manuscript

Orofacial motor functions in pediatric obstructive sleep apnea and implications for myofunctional therapy

Claudia Maria de Felício, Franciele Voltarelli da Silva Dias, Gislaine Aparecida Folha, Leila Azevedo de Almeida, Jaqueline Freitas de Souza, Wilma Terezinha Anselmo-Lima, Luciana Vitaliano Voi Trawitzki, Fabiana Cardoso Pereira Valera

PII: S0165-5876(16)30276-2

DOI: 10.1016/j.ijporl.2016.08.019

Reference: PEDOT 8222

To appear in: International Journal of Pediatric Otorhinolaryngology

Received Date: 6 May 2016

Revised Date: 22 August 2016 Accepted Date: 24 August 2016

Please cite this article as: C. Maria de Felício, F. Voltarelli da Silva Dias, G.A. Folha, L. Azevedo de Almeida, J. Freitas de Souza, W.T. Anselmo-Lima, L.V.V. Trawitzki, F.C.P. Valera, Orofacial motor functions in pediatric obstructive sleep apnea and implications for myofunctional therapy, *International Journal of Pediatric Otorhinolaryngology* (2016), doi: 10.1016/j.ijporl.2016.08.019.

This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.



Orofacial motor functions in pediatric obstructive sleep apnea and implications for myofunctional therapy

Claudia Maria de Felício^{1,2}
Franciele Voltarelli da Silva Dias^{1,2}
Gislaine Aparecida Folha^{1,2}
Leila Azevedo de Almeida³
Jaqueline Freitas de Souza^{1,2}
Wilma Terezinha Anselmo-Lima^{1,2}
Luciana Vitaliano Voi Trawitzki^{1,2}
Fabiana Cardoso Pereira Valera^{1,2}

- (1) Department of Ophthalmology, Otorhinolaryngology and Head and Neck Surgery, School of Medicine of Ribeirão Preto, University of São Paulo USP Ribeirão Preto (SP), Brazil.
- (2) Craniofacial Research Support Center, University of São Paulo USP Ribeirão Preto (SP), Brazil.
- (3) Department of Neurosciences and Behavioral Sciences, School of Medicine of Ribeirão Preto, University of São Paulo USP Ribeirão Preto (SP), Brazil.

Correspondence: Cláudia Maria de Felício, Departamento de Oftalmologia, Otorinolaringologia, e Cirurgia de Cabeça e Pescoço, Faculdade de Medicina de Ribeirão Preto, Universidade de São Paulo,

Av. dos Bandeirantes, 3900, Ribeirão Preto, SP, Brasil.

CEP: 14049-900

Tel: (+55 16) 33150592; Fax: (+55 16) 36022860 email: cfelicio@fmrp.usp.br

Conflicts of interest – The authors declare no conflicts of interest.

Abstract

Objectives: The purposes of this study were (1) to identify possible differences in muscular and orofacial functions between children with obstructive sleep apnea (OSA) and with primary snoring (PS); (2) to examine the standardized difference between normal values of myofunctional scores and those of subjects with OSA or PS; and (3) to identify the features associated with OSA.

Methods: Participants were 39 children (mean age 8 \pm 1.2 years) of which, 27 had a diagnosis of OSA and 12 had PS. All participants were examined by an otorhinolaryngologist and underwent overnight polysomnography. Orofacial characteristics were determined through a validated protocol of orofacial myofunctional evaluation with scores (OMES), surface electromyography of masticatory muscles, and measurements of maximal lip and tongue strength. Reference values in the OMES were included to quantify the standardized difference (effect size = ES) relative to the groups studied and in the regression analysis.

Results: The OSA group had lower scores in breathing and deglutition, more unbalanced masticatory muscle activities than PS group (P < 0.05), but both groups had similar reductions in orofacial strength. OSA had a large ES (Cohen's d > 0.8) in all analysed OMES scores, while PS group showed small and medium differences in breathing and mastication scores, respectively. The mobility of the stomatognathic components score was the most important to contribute for group status (57%, P < 0.0001) in the regression analysis.

Conclusion: Children with tonsillar hypertrophy and OSA had relevant impairments in orofacial functions and lesser muscular coordination than children with PS.

Keywords: obstructive sleep apnea, orofacial functions, electromyography, orofacial strength, myofunctional therapy, children

1. Introduction

Orofacial and pharyngeal muscles are involved in important functions including breathing, with the vital role of maintaining airflow. Any upper airway (UA) obstruction may induce changes in neuromuscular function in order to ensure the passage of air [1]. The most common consequence of UA obstruction is mouth breathing, a functional adaptation that may affect craniofacial growth and development during childhood [2]. Another possible consequence is obstructive sleep apnea (OSA) a sleep disordered breathing (SDB) that affects 1-5% of the children population [3].

Pediatric OSA is characterized by intermittent complete or partial obstruction (obstructive apnea or hypopnea) of UA, prolonged partial obstruction of UA, or both. This obstruction disrupts normal ventilation and influences on normal sleep patterns [4].

Increases in the size of tonsils and adenoids cause airway narrowing, and this is the most common factor associated with pediatric OSA. Thus, adenotonsillectomy (AT) is the first-line treatment for OSA during childhood, with reported cure or improvement of the disorder in most cases [5]. Nevertheless, the proportion of patients with postoperative residual OSA ranged from 13% to 29% in low-risk populations and reached 73% when obese children were included in the analysis [3]. AT can bring partial recovery of facial muscular and functional changes, particularly during the first month after surgery, but myofunctional disorders persist after six months [6].

Therefore, in addition to anatomic airway narrowing, neuromuscular factors should be considered as contributors to OSA, such as decreased upper airway dilator muscles function during sleep or poor muscle activation. Based on this assumption, some authors turned their focus to the relationship between neuromuscular factors and OSA, PS, or residual OSA after AT or orthodontic treatment in childhood, concluding that orofacial myofunctional therapy (OMT) should be included in the treatment of SDB in children [7-12].

Briefly, OMT is aimed at correcting abnormal breathing patterns and muscular dysfunctions that may impair upper airway patency [7]. Promising results such as decreases in the apnea-hypopnea index (AHI) have been described following an OMT program in adults with OSA [13] and children with residual OSA [9]. Therefore, OMT is proving to be valuable in the treatment of SDB, although relevant concerns have been raised in respect to the lack of clarity regarding the principles of neuromuscular rehabilitation [14], consensus about exercise types [11], and the specificity of therapeutic targets and procedures [15]. Moreover, muscular functions should be reevaluated after OMT in order to determine whether positive results in PSG are actually related to improvements in muscle and orofacial functions [15]. To our knowledge, only

Villa et al. (2015a) [9] described outcome measures such as breathing, nasal patency, and lip function to date.

The first step in an attempt to clarify these issues is to achieve a better understanding of the orofacial muscular profile of children with OSA. The use of a validated protocol for orofacial myofunctional evaluation and well-established measures of muscular strength and activity is fundamental in this effort. It is only after this step that the most suitable therapeutic strategies for the relief of conditions that contribute to OSA can be adequately planned.

In this study, we assessed children with OSA and PS in terms of their orofacial myofunctional characteristics (appearance/posture, mobility, and functions), activity and coordination of jaw muscles, and lip and tongue strength. Our aims were (1) to identify possible differences between children with OSA and PS; (2) to examine the standardized difference between normal myofunctional scores and those of subjects with OSA or PS; and (3) to identify the possible features associated with OSA.

2. Materials and Methods

This prospective study was approved by the local institutional ethics committee (process number 13214/2013) and informed consent was obtained from all parents for the anonymous use of their children's clinical data for research purposes.

A total of 39 children aged 7-10 years (mean \pm standard deviation = 8 \pm 1.2 years) were included in the study. From these, 27 (11 boys and 16 girls) were diagnosed with OSA and 12 (8 boys and 4 girls) with PS.

All participants were clinically evaluated by an otorhinolaryngologist and underwent a polysomnography test (PSG) at University Hospital. Oroscopy was used to assess the degree of tonsil hypertrophy based on the classification of Brodsky and Koch (1992) [16], and nasoendoscopy with a *Fujinon* flexible endoscope for children was used to evaluate adenoid hipertrophy.

Overnight PSG exams were performed using a *Biol-Logic*® digital polygraph and the software *SleepScan Vision*®, version 2.03.05. The technical parameters and sleep staging were performed according to the guidelines of the AASM [4]. During PSG exams, the following parameters were assessed: electroencephalogram (F3-M2, F4-M1, C3-M2, C4-M1, O1-M2, O2-M1), electrooculogram (E1-M2 and E2-M2), electrocardiogram (one derivation), electromyogram (submental area, tibialis anterior), nasal and oral airflow (thermistor), nasal pressure, chest and abdominal respiratory effort (inductance plethysmography), pulse oximeter, snoring (microphone), body

position, and synchronized video system. Analysis of these parameters observed the AASM recommendations [4]. All recordings were scored visually by an investigator who was blind in respect to the previous otorhinolaryngologic examination of the subjects.

Thus, based on the interview, examination, and PSG, these patients were allocated to one of two groups, OSA and PS.

The OSA group comprised subjects with symptoms suggestive of OSA, adenotonsillar hipertrophy (i.e. pharyngeal tonsil \geq 70% and palatine tonsils grade III or IV) [2], and a diagnosis of OSA in PSG [obstructive apnea-hypopnea index (OAHI) \geq 1.0 event/hour during sleep], according to the AASM [4].

The PS group consisted of 12 subjects with snoring complaints reported by parents and without adenotonsillar hypertrophy. During PSG, intermittent noise was verified in these patients, but without obstructive apnea or increased respiratory, and the OAHI was lower than 1.0 event/hour during sleep record.

Children with genetic syndromes, severe facial changes/malocclusion, neuromuscular diseases, body mass index (BMI) with Z-score above +2 above the reference values for age and sex of the World Health Organization (WHO), previous adenotonsillectomy or palate surgery, and previous or current orthodontic treatment or myofunctional therapy were not included in the study.

The sample distribution, age, body mass index (BMI) and OAHI are presented in Table 1.

2.1 Data Collection

First, the examiner instructed the subjects about the evaluations and procedures of the study. The participants were then evaluated while sitting on a chair with no headrest, with the head in natural position and feet on the ground, in a room with appropriate lighting. All evaluation/examination were performed by trained examiners who was blind in respect to the previous examination of the subjects.

2.1.1 Orofacial Evaluation

2.1.1.1 Orofacial Myofunctional Evaluation

Subjects were individually evaluated according to the protocol of the Orofacial Myofunctional Evaluation with Scores (OMES), proposed and validated for children

aged 6-12 years by Felício and Ferreira (2008) [17]. The evaluation was recorded for real time and future analysis [15, 17].

The OMES protocol has predetermined scores, with the highest values indicating normal patterns. Total score range from 32 to 104, with the highest value indicating the better orofacial myofunctional condition, and the lowest value the worse degree myofunctional disorder. The categories assessed and their respective items were the following:

- Appearance/Posture: face (symmetry), cheeks, mandible, lips, tongue, and hard palate.
- Mobility: subjects were asked to perform 4-6 movements with each component of lips, tongue, cheeks, or mandible.
 - Functions: breathing mode, deglutition and mastication.

This current version has scores of 4 to 1 for lips behavior during deglutition, aiming to exclude the score zero (of 3 to 0). Also, the item "bite" has been included in the mastication function, as compared to previous version [17]. The maximum possible score for functions was 29 (breathing = 3; deglutition = 16, and mastication = 10).

For additional support, please see Supplementary material.

A complementary part of the OMES protocol includes the analysis of occlusion with measures of the range of jaw movements, presence/absence of malocclusion, and pain and noise in the temporomandibular joint [17]. No scores are used in the complementary assessment.

2.1.1.2 Surface Electromyography (sEMG)

The masseter and anterior temporal muscles (left and right) were examined, during maximum voluntary clenching (MVC), following procedures described elsewhere [18-19].

All subjects underwent two tasks: (1) a standardization recording of MVC with two 10-mm thick cotton rolls positioned on the right and left mandibular second premolars/ first molars, and (2) a test recording during MVC in the intercuspal position. Each task had a duration of five seconds. After surface electromyographic (sEMG) potentials recording, two EMG indices, based on those proposed by Naeije et al. (1989) [20] were calculated. Although the EMG potentials recorded during the MVC tests were expressed as percentage of the mean potentials recorded during MVC on the cotton rolls (unit: μ V/ μ V x 100), in other words, standardized potentials, as recommended by Ferrario et al. (2000) [18]. The calculated indices were:

Asymmetry (unit: %), which refers to the symmetric/asymmetric activity of the right and left masticatory muscles.

Activity (unit: %), which shows whether one of muscle pairs (temporalis or masseters), was prevalent or not during clenching. Positive values of standardized potentials indicates the prevalence of masseter muscles, whereas negative values reflect the prevalence of temporalis muscles.

EMG activity was recorded using a wireless electromyographic system (FreeEMG, BTS S.p.A., Garbagnate Milanese, Italy) and for calculations, the SMART Analyzer software (BTS S.p.A.) was used.

The EMG indices reproducibility and the Technical Error of Measurement (random error) were previously tested in our laboratory [19].

2.1.1.3 Orofacial Strength

Maximal lip and tongue strength were measured with the *Iowa Oral Performance Instrument* (IOPI), model 2.2. (IOPI MedicalLLC – Redmond, WA - USA). The IOPI measures the pressure in kilopascals (kPa) that an individual can produce by pressing a standardized air-filled tongue bulb. All participants practiced the tasks before the measurements were recorded.

The lip task was performed with the IOPI bulb sandwiched between two wooden tongue blades and positioned between the lips at midline. Participants were instructed to maintain the teeth occluded and to separate and protrude the lips slightly as the blades were positioned. They were then asked to press the lips together with maximum effort [21].

In respect to tongue strength, three parameters were measured: anterior tongue elevation, deglutition, and tongue protrusion. Anterior tongue elevation was measured with the bulb positioned immediately posterior to the central incisors, while the bulb stem was held by the examiner immediately before the central incisors. The children were asked to raise their tongues and squeeze the bulb against the palate as hard as they could for approximately 3s [22]. During this task, participants were encouraged to rest their incisors gently on the tubing of the IOPI bulb [21]. Deglutition was measured with the bulb positioned as described above and participants were asked to swallow saliva as usual.

Finally, tongue protrusion was measured with the bulb attached to a bulb-holder and positioned between the upper and lower incisors, with the tongue bulb facing the inside of the mouth. Participants were instructed to protrude the tongue as hard as possible against the bulb, which was held firmly in place (via adapter) by the teeth [21].

Three strength measures were acquired in each test, with a resting period of about 30s between trials. The highest pressure across the three trials was used as the participants' maximal isometric pressure [23].

2.1.2 Examiner

A speech-language pathologist, previously trained and with good reliability indicators, performed all the evaluations.

2.1.3 Reference Sample

Reference values in the OMES of healthy children (n = 15, age range: of 6 - 11 years, mean age = 9 \pm 1.6) with total OMES score above the cut-off value of 85 (indicating absence of relevant orofacial myofunction disorder) assessed in a previous study [24], was included for comparison with groups OSA and PS (likely a priori meta-analyses). For this purpose, hard palate scores were not included in the analysis because data from the reference group were collected from video images that preclude the analysis of this item. Therefore, the maximum possible OMES score was 101 instead of 104.

2.1.4 Data Analysis

Descriptive statistics were computed for all variables and are expressed as mean and standard deviation (SD). The technical error of measurement (random error) [19] was computed for two repeated measures (trials 2 and 3) of the lip and tongue strength tasks.

The non-parametric Mann-Whitney test was used for comparative analyses between groups with OSA and PS. Data from the reference sample were used to estimate the effect size (ES) of PS or OSA on myofunctional conditions in relation to normal values, as well as to check for associations between group status and categories of the OMES protocol (regression analysis), as explained below.

The ES was computed by using the group means and adjusting the pooled standard deviations with weights for the sample sizes because of the different numbers of subjects in the groups. The ES refers to the magnitude of the difference between a given measure and a standardized measure (Z) and can be interpreted as small (d = 0.2), medium (d = 0.5), and large (d = 0.8) according to Cohen (1977) [25].

For univariate and multiple regression analyses, group status was treated as a measure with values of 1 for OSA, 2 for PS and 3 for reference sample. We chose this simple model, with an equal distance between statuses, in the absence of evidence for a more complex model to describe myofunctional conditions in different groups. Univariate analysis was used to select variables eligible (P < 0.2), to enter the multiple regression analysis which was used to estimate those categories of the OMES protocol (summary score of appearance/posture, mobility and functions) that best predicted changes in the dependent variable (group status).

The analyses were made with *Statistica 13* (Dell software Inc., Aliso Viejo, United States of America). The level of statistical significance was set at P < 0.05. Effect sizes were measured with an online calculator available at http://www.psychometrica.de/effect size.html (accessed on 12/15/2015).

3. Results

3.1 Data reproducibility

For all the tongue and lip tasks of the IOPI, the test-retest random error was much lower than the intragroup standard deviation, showing the good reproducibility of the measures.

3.2 Orofacial Myofunctional Evaluation (OMES protocol)

3.2.1 OSA and PS groups comparison

Participants had a normal range of jaw movements and no muscular pain or temporomandibular joint pain or noise. Eight children in the OSA group, but none in PS group, had posterior crossbite. None of the subjects had abnormal tongue frenulum.

The OSA group had lower scores in breathing, swallowing, and in the functions summary score (P < 0.05) compared to the PS group. During deglutition, few subjects in each group (15-16%) had normal lip behavior, although the most pronounced alterations were seen in the OSA group. Examples of these changes were absence of anterior sealing of the oral cavity or excessive contraction of perioral muscles, with participation of the mentalis muscle to achieve lip closure. There was no significant difference between groups in the categories appearance/posture and mobility. Detailed results are shown in Table 2.

3.2.2 Effect Size

Subjects with OSA and PS had lower standardized scores compared to the reference group. In the OSA group, the ES was large for appearance/posture, mobility, breathing, swallowing, mastication, summary score of the functions, and OMES total score. The ES between the PS group and the reference group was small for breathing, medium for mastication, and large for the other comparisons (see Table 2).

3.2.3 Association Analysis

The univariate analysis showed that mobility and functions categories had P < 0.2. These variables were thus included in the multiple regression analysis, while category appearance/posture was not (P > 0.2). The multiple regression analysis showed that the category mobility was significantly associated with group status. There was no significant association between group status and functions. The multiple regression model explained 55% of the group status (P < 0.0001). The relative contribution (Beta coefficient) of the category mobility was 57% and the relative contribution of functions was 22% for the prediction of the dependent variable. Thus, mobility was the main category to explain group status.

3.3 Surface Electromyography (sEMG)

The results of sEMG of masticatory muscles during MVC showed that patients with OSA had significantly higher asymmetry between right and left muscles and higher activity index (the negative activity index indicates a prevalence of temporalis muscle over masseter) compared to subjects with PS. The means of muscle activity during MVC with cotton rolls (not standardized) in the OSA group were not significantly different from those of the PS group (Figures 1A and 1B).

3.4 Orofacial Strength

There were no significant differences across groups in terms of lip or tongue pressure (P > 0.05), as shown in Figure 2.

4. Discussion

The purpose of this study was to provide a better understanding of the orofacial myofunctional characteristics of children with SDB, especially OSA. Thus, a valid orofacial myofunctional evaluation protocol was adopted, associated with reproducible and reliable measures of muscle strength, activity, and coordination.

We found that (1) children with SDB present changes in orofacial myofunctional conditions, especially in the most severe group (OSA group); (2) orofacial mobility was the main distinctive feature between subjects with SDB and healthy reference subjects; and (3) coordination between masticatory muscles was lower in children with OSA compared to children with PS.

During the orofacial myofunctional evaluation, both the PS and OSA groups presented alterations in appearance/posture and mobility of the stomatognathic system components, with large standardized differences from the reference group. Most of the changes observed are consistent with data from studies with children with mouth breathing [2,6] and OSA [7,9,12]. However, the OSA group had significantly lower mean breathing scores, commonly found when mouth breathing is predominant, as compared to the PS group, with a large standardized difference (ES = 1.12) relative to the reference group. This result was expected and is likely related to the fact that all subjects with OSA had important adenotonsillar hypertrophy [6], in contrast to none in the PS group. As verified in previous studies, daytime mouth breathing (or mouth open during day) is one of the few items of the clinical history with significant difference between OSA e PS [26,27]. Nevertheless, not all participants in the PS group presented lip closure during the evaluation (41.67%), resulting in breathing scores with small standardized differences from the reference group.

Stomatognathic functions are determined by use and sensory experience, and mouth breathing is a factor that alters orofacial behaviors for the maintenance of the vital function. Mouth breathing requires changes in the position of lips, jaw, and tongue [2,6,28]. In this breathing mode, the lips are not sealed, the jaw is opened by the suprahyoid muscles and this displacement is followed by tongue. Moreover, when the palatine tonsil is hypertrophic, genioglossus muscle contraction moves the base of the tongue forward increasing the oropharyngeal space, at least during wakefulness. The persistence of the problem results in a chain of events, including changes in sensory inputs from the face and mouth and in neuromuscular function that precede the adaptation of craniofacial morphology [1].

Together, all these aspects may have caused the slightly increased impairment in swallowing in the OSA group relative to the PS group, regardless of both having similar tongue and lip strength. In both groups, means were lower than those reported for healthy children in the tongue elevation [22] and lip compression tasks [29]. In relation

to the other tongue tasks, we were unable to find normality values based on the same measurement method for the age range.

Despite the absence of difference between groups PS and OSA in mastication scores, the standardized difference relative to the reference group were medium and large, respectively. Factors underlying poor mastication scores may include muscular deficits, malocclusion, or both. Unilateral mastication is common during the period of mixed dentition due to natural occlusal instability, but sometimes it may become chronic. Therefore, the crossbite seen in subjects with OSA (30%) is relevant and should be referred for orthodontic treatment [10] for a better muscular and functional outcome.

According to the sEMG analysis, the studied groups had a similar capacity for maximal voluntary contraction of masticatory muscles (non-standardized potentials). However, the OSA group presented higher asymmetry between muscles of the right and left sides and dominance of the temporalis over masseter muscles during clenching, while the PS group had a more balanced distribution of muscle activity between sides and between muscle pairs. These indices provide a better estimation of the coordination of jaw muscle contraction than just sEMG potential amplitude, as previously defined [18,20]. The muscle asymmetry found in OSA may be linked to presence of crossbite in a portion of subjects in this group.

The first step in the treatment of pediatric OSA is adenotonsillectomy (AT) [3,5] followed by orthodontic treatment if necessary [10]. However, as maladapted orofacial functions may be irreversible or present insufficient improvement even when their original cause is eliminated [6], muscle and functional reeducation has been recommended [7-12].

The findings of our study show that the differences between the OSA group and the PS group were UA narrowing due to tonsil hypertrophy, from an anatomical point of view, and impaired breathing, worse deglutition patterns, and wore balance between masticatory muscles, from a functional point of view. Moreover, among the categories of the OMES protocol, mobility appeared as the determinant of group status (PS, OSA and reference group). Therefore, functional maladaptation caused by UA obstruction seems to be responsible for impaired orofacial motor control.

Orofacial functions involve the capacity for muscle contraction, but also a motor skill to coordinate agonist and antagonist muscles with adequate amplitude, in addition to speed control and accuracy of movement in accordance with the task demands.

Swallowing and mastication, which are semiautomatic functions, as well as voluntary movements like those employed in mobility tasks, are under control of the primary motor area representing the orofacial muscles (face-M1). Face-M1 operates in

integration with other cortical and subcortical regions [30] and employs somatosensory inputs from the face and mouth, also playing a role in adaptive processes with reorganization of representations or changes in face-M1 excitability (i.e., neuroplasticity) [31]. Neuroplasticity may also reflect behavioral maladaptation in some instances, such as in oral alterations and orofacial pain [30].

Because this sophisticate system is also involved in the learning of novel oral motor tasks, researchers have drawn attention to the relevance of the principles of neural plasticity for the development of new treatment strategies for masticatory, orofacial, and oropharyngeal impairments [30,32].

Researchers have successfully tested the neuroplastic effects of learning and repetition involved in a tongue task (not oriented for strength gain) on corticomotor excitability [33,34]. Recently, authors investigated whether an improvement in the coordination of the tongue in a protrusion task (Genioglossus Muscle, GG, Force= 1.0 N) could decrease obstructive breathing disturbances during REM sleep. The positive results found were attributed to an improvement in UA stability due to enhancement in GG cortical excitability [35].

Our results should be interpreted with caution because of the limited number of participants and the lack of reference data (normal values) for other variables besides myofunctional condition that could be compared to those of the samples studied. Moreover, similarities in behavioral and health problems has been demonstrated between children with PS and OSA [36]. Further investigation is therefore necessary, including larger samples and participants without breathing disturbances.

The therapeutic implications of these findings is that children with SDB require myofunctional intervention to promote nasal breathing, orofacial muscle strength and adequate posture of stomatognathic system components, as proposed [7-12]. Moreover, especially children with OSA would benefit from strategies to improve orofacial motor control.

In conclusion, children with OSA had more impaired breathing mode, deglutition pattern and coordination of masticatory muscles than PS. The greatest alterations of orofacial appearance/posture, mobility and functions were found in OSA group, as compared to reference group. Further studies are needed to explore the possibility of orofacial motor control being involved in children with OSA.

5. Acknowledgements:

Financial support: Provost's Office for Research of the University of São Paulo (Process No. 11.1.21626.01.7).

The third author received a fellowship from the Coordination of Improvement of Higher Education Personnel (CAPES), Brazil.

6. References

- [1] A.J. Miller, K. Vargervik, G. Chierici, Experimentally induced neuromuscular changes during and after nasal airway obstruction, Am. J. Orthod. 85 (1984) 385-392.
- [2] F.C. Valera, L.V. Travitzki, S.E. Mattar, M.A. Matsumoto, A.M. Elias, W.T. Anselmo-Lima, Muscular, functional and orthodontic changes in preschool children with enlarged adenoids and tonsils, Int. J. Pediatr. Otorhinolaryngol. 67 (2003) 761-770.
- [3] C.L. Marcus, L.J. Brooks, K.A. Draper, D. Gozal, A.C. Halbower, J. Jones, et al., Diagnosis and management of childhood obstructive sleep apnea syndrome, Pediatrics. 130 (2012) e714-755.
- [4] American Academy of Sleep Medicine, International classification of sleep disorders, third ed., Darien, Illinois, 2012.
- [5] C.L. Marcus, R.H. Moore, C.L. Rosen, B. Giordani, S.L. Garetz, H.G. Taylor, et al., Childhood Adenotonsillectomy Trial (CHAT), A randomized trial of adenotonsillectomy for childhood sleep apnea, N. Engl. J. Med. 368 (2013) 2366-2376.
- [6] D.A. Bueno, T.H. Grechi, L.V. Trawitzki, W.T. Anselmo-Lima, C.M. Felício, F.C. Valera, Muscular and functional changes following adenotonsillectomy in children, Int. J. Pediatr. Otorhinolaryngol. 79 (2015) 537-540.
- [7] C. Guilleminault, Y.S. Huang, P.J. Monteyrol, R. Sato, S. Quo, C.H. Lin, Critical role of myofascial reeducation in pediatric sleep-disordered breathing, Sleep Med. 14 (2013) 518-525.
- [8] J. Moeller, L.C. Paskay, M.L. Gelb, Myofunctional therapy: a novel treatment of pediatric sleep-disordered breathing, Sleep Med. Clin. 9 (2014) 235-243.
- [9] M.P. Villa, L. Brasili, A. Ferretti, O. Vitelli, J. Rabasco, A.R. Mazzotta, et al., Oropharyngeal exercises to reduce symptoms of OSA after AT, Sleep Breath. 19 (2015a) 281-289.
- [10] M.P. Villa, A. Rizzoli, J. Rabasco, O. Vitelli, N. Pietropaoli, M. Cecili, et al., Rapid maxillary expansion outcomes in treatment of obstructive sleep apnea in children, Sleep Med. 16 (2015b) 709-716.
- [11] M. Camacho, V. Certal, J. Abdullatif, S. Zaghi, C.M. Ruoff, R. Capasso, et al., Myofunctional therapy to treat obstructive sleep apnea: a systematic review and meta-analysis, Sleep. 38 (2015) 669-675.

- [12] S.Y. Lee, C. Guilleminault, H.Y. Chiu, S.S. Sullivan, Mouth breathing, "nasaldisuse," and pediatric sleep-disordered breathing, Sleep Breath. 19 (2015) 1257-1264.
- [13] K.C. Guimarães, L.F. Drager, P.R. Genta, B.F. Marcondes, G. Lorenzi-Filho, Effects of oropharyngeal exercises on patients with moderate obstructive sleep apnea syndrome, Am. J. Respir. Crit. Care Med. 179 (2009) 962-966.
- [14] C.M. Steele, On the plausibility of upper airway remodeling as an outcome of orofacial exercise, Am. J. Respir. Crit. Care Med. 179 (2009) 858-859.
- [15] G.A. Folha, F.C. Valera, C.M. de Felício, Validity and reliability of a protocol of orofacial myofunctional evaluation for patients with obstructive sleep apnea, Eur. J. Oral Sci. 123 (2015) 165-172.
- [16] L. Brodsky, R.J. Koch, Anatomic correlates of normal and diseased adenoids in children, Laryngoscope. 102 (1992) 1268-1274.
- [17] C.M. Felício, C.L. Ferreira, Protocol of orofacial myofunctional evaluation with scores, Int. J. Pediatr. Otorhinolaryngol. 72 (2008) 367-375.
- [18] V.F. Ferrario, C. Sforza, A. Colombo, V. Ciusa, An electromyographic investigation of masticatory muscles symmetry in normo-occlusion subjects, J. Oral Rehabil. 27 (2000) 33-40.
- [19] C.M. De Felício, C.L. Ferreira, A.P. Medeiros, M.A. Rodrigues Da Silva, G.M. Tartaglia, C. Sforza, Electromyographic indices, orofacial myofunctional status and temporomandibular disorders severity: a correlation study, J. Electromyogr. Kinesiol. 22 (2012) 266-272.
- [20] M. Naeije, R.S. McCarroll, W.A. Weijs, Electromyographic activity of the human masticatory muscles during submaximal clenching in the inter-cuspal position, J. Oral Rehabil. 16 (1989) 63-70.
- [21] H.M. Clark, N.P. Solomon, Age and sex differences in orofacial strength, Dysphagia. 27 (2012) 2-9.
- [22] N.L. Potter, R. Short, Maximal tongue strength in typically developing children and adolescents, Dysphagia. 24 (2009) 391-397.
- [23] J. Vanderwegen, C. Guns, G. Van Nuffelen, R. Elen, M. De Bodt, The influence of age, sex, bulb position, visual feedback, and the order of testing on maximum anterior and posterior tongue strength and endurance in healthy belgian adults, Dysphagia. 28 (2013) 159-166.
- [24] C.M. De Felício, G.A. Folha, C.L.P. Ferreira, A.P. Medeiros, Expanded protocol of orofacial myofunctional evaluation with scores: validity and reliability, Int. J. Pediatr. Otorhinolaryngol. 74 (2010) 1230-1239.

- [25] J. Cohen, Statistical power analysis for the behavioral sciences, Revised ed. Academic Press, New York, 1977, pp.1-17.
- [26] J.L. Carroll, S.A. McColley, C.L. Marcus, S. Curtis, G.M. Loughlin, Inability of clinical history to distinguish primary snoring from obstructive sleep apnea syndrome in children, Chest. 108 (1995) 610-618.
- [27] K. Bertran, T. Mesa, K. Rosso, M.J. Krakowiak, E. Pincheira, P.E. Brockmann, Diagnostic accuracy of the spanish version of the pediatric sleep questionnaire for screening of obstructive sleep apnea in habitually snoring children. Sleep Med. 16 (2015) 631-636.
- [28] D.B. Berlese, P.F.F. Fontana, L. Botton, A.R.M. Weimnan, L.S.B. Haeffner, Myofunctional characteristics of obese mouth and nose breathers. www.scielo.br/pdf/rsbf/v17n2/en_12.pdf (accessed 19.08.16).
- [29] K.M. Van Lierde, K. Bettens, A. Luyten, J. Plettinck, K. Bonte, H. Vermeersch, et al., Oral strength in subjects with a unilateral cleft lip and palate, Int. J. Pediatr. Otorhinolaryngol. 78 (2014) 1306-1310.
- [30] L. Avivi-Arber, R. Martin, J.C. Lee, B.J. Sessle, Face sensorimotor cortex and its neuroplasticity related to orofacial sensorimotor functions, Arch. Oral Biol. 56 (2011) 1440-1465.
- [31] L. Avivi-Arber, J.C. Lee, B.J. Sessle, Dental occlusal changes induce motor cortex neuroplasticity, J. Dent. Res. 94 (2015) 1757-1764.
- [32] J. Robbins, S.G. Butler, S.K. Daniels, R. Diez Gross, S. Langmore, C.L. Lazarus, et al., Swallowing and dysphagia rehabilitation: translating principles of neural plasticity into clinically oriented evidence, J. Speech Lang. Hear. Res. 51 (2008) S276-S300.
- [33] P. Svensson, A. Romaniello, K. Wang, L. Arendt-Nielsen, B.J. Sessle, One hour of tongue-task training is associated with plasticity in corticomotor control of the human tongue musculature, Exp. Brain Res. 173 (2006) 165-173.
- [34] Y. Komoda, T. Iida, M. Kothari, O. Komiyama, L. Baad-Hansen, M. Kawara, et al., Repeated tongue lift movement induces neuroplasticity in corticomotor control of tongue and jaw muscles in humans, Brain Res. 1627 (2015) 70-79.
- [35] E, Rousseau, C. Silva, S. Gakwaya, F. Sériès, Effects of one-week tongue task training on sleep apnea severity: a pilot study, Can. Respir. J. 22 (2015) 176-178.
- [36] A.M. Zicari, F. Occasi, F. Di Mauro, V. Lollobrigida, M. Di Fraia, V. Savastano, et al., Mean platelet volume, vitamin D and C reactive protein levels in normal weight children with primary snoring and obstructive sleep apnea syndrome. PLoS One. 11 (2016) e0152497.

Table 1: Characteristics of subjects with primary snoring (PS) and obstructive sleep apnea (OSA) evaluated in the study. Median, Interquartil Range (IQR), Mean and Standard Deviation (SD)

	PS (n=12)				OSA (n=27)				P
Variables	Median	IQR	Mean	SD	Median	IQR	Mean	SD	
Age (years)	7.50	2.00	7.92	1.24	8.00	1.00	7.70	1.23	0.64
BMI (Kg/m²)	15.50	2.21	15.80	2.00	15.91	2.82	16.65	1.90	0.24
OAHI (e/h)	0.70	0.45	0.60	0.29	2.2	1.80	3.08	2.22	<0.001

P: probability of Mann-Withney test. *P*< 0.05 difference statistically significant. BMI: body mass index; OAHI: Obstructive apnea-hypopnea index; e/h event/hour

Table 2: Orofacial myofunctional scores on OMES-protocol of the Reference (R), PS and OSA groups. Mean, standard deviation (SD) and Effect size (ES)

and OSA groups. Mean	Refere		PS		OSA		
	(N= '	15)	(N=12	2)	(N= 27)		
	Mean	SD	Mean	SD	Mean	SD	Р
Appearance/posture*	12.60	1.35	10.92	1.68	10.81	1.92	0.71
ES (95%CI)			1.12(1.9	3/0.30)	1.03(1.67/0.36)		
Mobility score	52.93	2.52	46.25	3.19	43.85	4.14	0.10
ES (95%CI)			2.35 (3.34/1.31)		2.46(3.38/1.63)		
Functions							
Breathing	2.80	0.41	2.58	0.51	2.11	0.70	0.048
ES (95%CI)			0.48 (1.25/0.28)		1.12(1.8/0.45)		
Deglutition	15.13	0.83	13.75	1.06	12.59	1.89	0.047
ES (95%CI)			1.44 (2.36/0.62)		1.59(2.30/0.87)		
Mastication	8.47	1.30	7.75	1.29	7.07	1.59	0.19
ES (95%CI)			0.61 (1.39/0.17)		0.94(1.59/0.27)		
Summary Score	26.4	1.73	24.08	2.19	21.78	3.14	0.045
Functions							
ES (95%CI)			1.20 (2.02/0.37)		1.69(2.42/0.97)		
OMES-score*	91.93	3.41	81.25	6.11	76.44	7.50	0.067
ES (95%CI)			2.23 (3.19/1.26)		2.43(3.25/1.61)		

^{*} Appearance/posture and OMES-score were calculated without hard palate score.

Effect size (Cohen's *d*), CI: confidence limits, P: probability of Mann-Withney test. P< 0.05 difference statistically significant, R: Reference group (subjects with normal OMES-score higher than the cut-off value of 85).

Captions for Figures

Figure 1: (A) Potentials sEMG of the masticatory muscles during the MCV with cotton rolls. (B) Standardized sEMG indices asymmetry and activity (normal range -10% to 10%) of the obstructive sleep apnea (OSA) and PS (primary snoring) groups. Median, 1th and 3th quartile, minimum, maximum and outside values. Mann-Withney test, P < 0.05: significant difference, ns: not significant.

Figure 2. Orofacial strength: Lips and tongue pressure of the obstructive sleep apnea (OSA) and PS (primary snoring) groups. Median, 1th and 3th quartile, minimum, maximum and outside values. Mann-Withney test, ns: not significant (P > 0.05).





