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PEDIATRICS • ORIGINAL ARTICLE

Can myofunctional therapy increase tongue tone and reduce symptoms in children with sleep-disordered breathing?

Maria Pia Villa[1] · **Melania Evangelisti**[1] · **Susy Martella**[1] · **Mario Barreto**[1] · **Marco Del Pozzo**[1]

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Abstract
Purpose Data in the literature suggest that myofunctional therapy (MT) may be able to play a role in the treatment of children with sleep-disordered breathing (SDB). Our study investigated the effectiveness of MT in reducing respiratory symptoms in children with SDB by modifying tongue tone.
Methods Polysomnographic recordings were performed at baseline to assess obstructive sleep apnea (OSA) severity in 54 children (mean age 7.1 ± 2.5 years, 29 male) with SDB. Patients were randomly assigned to either the MT or no-MT group. Myofunctional evaluation tests, an assessment of tongue strength, tongue peak pressure, and endurance using the Iowa Oral Performance Instrument (IOPI), and nocturnal pulse oximetry were performed before (T0) and after (T1) 2 months of treatment.
Results MT reduced oral breathing (83.3 vs 16.6%, $p < 0.0002$) and lip hypotonia (78 vs 33.3%, $p < 0.003$), restored normal tongue resting position (5.6 vs 33.4%, $p < 0.04$), and significantly increased mean tongue strength (31.9 ± 10.8 vs 38.8 ± 8.3, $p = 0.000$), tongue peak pressure (34.2 ± 10.2 vs 38.1 ± 7.0, $p = 0.000$), and endurance (28.1 ± 8.9 vs 33.1 ± 8.7, $p = 0.01$) in children with SDB. Moreover, mean oxygen saturation increased (96.4 ± 0.6 vs 97.4 ± 0.7, $p = 0.000$) and the oxygen desaturation index decreased (5.9 ± 2.3 vs 3.6 ± 1.8, $p = 0.001$) after MT.
Conclusions Oropharyngeal exercises appear to effectively modify tongue tone, reduce SDB symptoms and oral breathing, and increase oxygen saturation, and may thus play a role in the treatment of SDB.

Keywords Children · Obstructive sleep apnea · Myofunctional therapy · Oropharyngeal exercises

Introduction

Sleep-disordered breathing (SDB) is an upper airway dysfunction that occurs during sleep and is characterized by snoring and/or a greater respiratory effort caused by increased upper airway resistance and pharyngeal collapsibility [1].

Obstructive sleep apnea (OSA) is the most severe clinical type of SDB, and the most common cause of OSA is adenotonsillar hypertrophy, though other anatomical and neuromuscular factors such as craniofacial dysmorphism, obesity, and hypotonic neuromuscular disease are also involved [2, 3].

The multidisciplinary therapeutic approach to OSA is based on adenotonsillectomy, orthodontic and medical treatments, weight loss, and non-invasive ventilation [1, 4, 5]. Treatment interventions are applied in a stepwise fashion until the complete resolution of SDB is achieved. Different treatment modalities are often combined depending on the severity and underlying conditions predisposing to upper airway obstruction during sleep [1, 4, 5]. Oral breathing and lip hypotonia, which are peculiar characteristics of children with OSA, increase nasal resistance and are associated with malposition of the tongue, thereby exacerbating the impaired development and further hampering the growth of the maxilla and mandible [6–8]. Persistence of oral breathing during sleep directly affects the position and strength of the tongue as well as that of the orofacial muscles, thereby causing abnormal airway

✉ Maria Pia Villa
mariapia.villa@uniroma1.it

[1] Pediatric Unit Sant'Andrea Hospital, NESMOS Department, Faculty of Medicine and Psychology, "Sapienza" University, Rome, Italy

development and SDB [8]. Myofunctional re-education is performed to avoid this evolution and may prove useful as a means of restoring a correct stomatognathic function and of treating OSA associated with other treatments so as to avoid residual OSA [9, 10].

The aims of our study were to evaluate the efficacy of myofunctional therapy (MT) as a first step designed to reduce oral breathing in children with SDB and to evaluate the increase in tongue tone using the Iowa Oral Performance Instrument (IOPI).

Methods

This prospective, case–control study was based on children who were referred to our Pediatric Sleep Center (S. Andrea Hospital, Rome, Italy) for SDB. The diagnosis of OSA was confirmed by the presence of SDB symptoms combined with a polysomnography (PSG) yielding an obstructive apnea/hypopnea index (AHI) >1 event/h [1].

Exclusion criteria were a positive history of acute or chronic cardiorespiratory or neuromuscular diseases, chronic inflammatory diseases, major craniofacial abnormalities, chromosomal syndromes, and epilepsy.

Age- and sex-matched control healthy Caucasian children were randomly recruited from a school in the same urban area of the study group. The inclusion criterion was as follows: none of the controls was obese. No history of sleep problems (snoring, apnoeas, and restless sleep) were reported. The control group underwent only IOPI measurements.

A clinical examination was performed and a sleep clinical record (SCR) obtained for all the SDB patients.

Polysomnographic recordings were performed before MT to define OSA severity. Thereafter, patients with SDB were randomized to two groups—an SDB case group (MT group) treated with MT plus nasal washing, consisting of the application of a saline solution in each nostril three times a day, and an SDB control group (no-MT group) treated with nasal washing alone. We measured tongue strength, tongue peak pressure, and endurance using the IOPI, and performed nocturnal pulse oximetry before (T0) and after (T1) 2 months of treatment in all the patients. We compared the baseline IOPI measurements of the SDB groups with normal values obtained from 38 sex- and age-matched, healthy school children.

The "Sapienza" University institutional review board approved the study, and written informed consent was obtained from the parents of the children upon admission to the study.

Sleep clinical record

All the children underwent the SCR, which has been validated previously [11]. Briefly, the SCR consists of three main sections. The first section takes into consideration the data yielded by a physical examination of the nose, oropharynx, and dental and skeletal occlusion. The following signs are considered: signs of oral breathing; nasal obstruction, inferior turbinate hypertrophy, or rhinolalia; visual evaluation of soft-palate position according to Friedman classes (grades 3 and 4 considered as positive); nasal septum deviation; tonsillar hypertrophy, with grades 3 and 4 being considered as tonsillar hypertrophy [12]; obese or adenoid phenotype; dental/skeletal malocclusion, and narrow hard palate. The dental/skeletal malocclusion, i.e., the intermaxillary divergence, includes jaw deviation from normal occlusion such as retrognathia; prognathia; open, deep bite; crossbite; and overjet. These signs were scored as either two points (positive sign) or zero point (negative sign).

The second section describes the patients' subjective symptoms as summarized by the Brouillette OSAS score. Questions investigated sleep symptoms (habitual snoring, witnessed apnoeic episodes, frequent awakenings, or agitated sleep). Questions were kept simple and concise, and a yes/no response format was chosen. We calculated the Brouillette score, considering a score equal to or higher than −1 as positive [13] and scoring it as 0.5. The third section consists in assessing the presence of inattention and hyperactivity symptoms using the attention deficit hyperactive disorder (ADHD) rating scale for school-aged children [14]. A score higher than 6 was considered as positive and scored as 1. A total sleep clinical record score of 6.5 or more was considered to be positive, as has previously been approved [11].

Sleep analysis

All the children underwent a laboratory overnight PSG assessment, using a Grass Heritage polygraph, in our Pediatric Sleep Center, to define the severity of OSA. The variables recorded included an electroencephalogram with at least eight channels (frontal, central, temporal, and occipital, referred to the contralateral mastoid), an electro-oculogram, a submental electromyogram, and an electrocardiogram (one derivation); chest and abdomen movements were measured by strain gauges. Airflow was recorded from an oronasal thermocouple and a nasal pressure transducer. Arterial oxygen saturation was monitored using pulse oximetry. Sleep stages were scored according to the standard criteria of the American Academy of Sleep Medicine (AASM). Arousals were detected visually according to the criteria reported in the recent manual for the scoring of sleep and associated events by the AASM [15].

Central, obstructive, and mixed apnoea events were counted according to the criteria established by the AASM [15]. The AHI was defined as the average number of apnoea and hypopnea events per hour of sleep. The diagnosis of OSA was confirmed if the laboratory PSG yielded an AHI ≥1 event/h combined with SDB symptoms [1].

Nocturnal pulse oximetry

The Nonin 2500A pulse oximeter (Nonin Medical; Plymouth, MN) was used to perform nocturnal oximetry recordings. A physician analyzed the recordings using the PROXYnet 10.1 software package (MedicAir, Italia). In brief, oximetry recordings with at least three clusters of desaturation events and at least three dips in SaO_2 below 90% were regarded as being diagnostic for OSA (abnormal or positive oximetry). Recordings not meeting these diagnostic criteria were considered as negative/inconclusive for OSA [16].

Myofunctional evaluation and treatment

The myofunctional therapist filled in a myofunctional evaluation form that assessed the respiratory pattern (nasal or oral), labial seal (competent or not), lip tone (normal or not), and tongue position at rest and during swallowing (normal or not normal) [17, 18].

The MT consisted of isometric and isotonic exercises involving the tongue, soft palate, and lateral pharyngeal wall designed to improve suction, swallowing, chewing, breathing, and speech functions.

Oropharyngeal exercises were divided into three categories: (1) nasal breathing rehabilitation, (2) labial seal and lip tone exercises, and (3) tongue posture exercises. The types of exercises used were described in a previous article [10].

Children were required to perform the exercises every day at home, at least three times a day, doing 10–20 repetitions each time. The MT group underwent two monthly meetings with a myofunctional therapist. In the first meeting (T0), the therapist carried out a myofunctional evaluation and taught the patients and their parents how to perform the rehabilitation exercises, which children were required to perform daily at home for 2 months.

In order to reduce a possible observer bias, one therapist performed all the myofunctional evaluations in both groups at T0 and T1, while another taught the children and their parents how to perform the exercises.

Nasal washing was performed using Rinowash with 2.5% saline hypertonic solution. All the patients performed nasal washing twice daily, in the morning and evening, for 2 months.

IOPI measurements

The IOPI objectively measures tongue and lip strength and endurance. Tongue strength is assessed by measuring the maximum pressure exerted when an individual presses a disposable, standard-sized tongue bulb against the roof of the mouth. The peak pressure achieved is displayed on a large, easy-to-read LCD. The units displayed are kilopascals (kPa), according to the internationally recognized unit of pressure, the pascal (Pa). The standard procedure and standard values

Table 1 PSG parameters and myofunctional evaluation of the two groups of children with SDB

	MT group 36)	No-MT group (18)	*p* value
Total sleep time (min)	440.5 ± 71.1	452.6 ± 67.7	0.8[a]
N1 NREM sleep (%)	13.6 (7.7–18.9)	13.5 (10.3–23.3)	0.9[a]
N2 NREM sleep (%)	40.5 (32.9–49.4)	34.2 (29.4–44.0)	0.2[a]
N3 NREM sleep (%)	26.1 (16.9–30.0)	28.7 (21.9–35.1)	0.2[a]
REM (%)	15.4 (9.8–18.6)	19.0 (12.3–22.5)	0.2[a]
Arousal index (*n*/h)	0.0 (0.0–4.5)	0.0 (0.0–3.2)	0.6[a]
AHI (ev/h)	1.5 (1.0–2.8)	1.8 (1.0–2.5)	0.9[a]
SpO_2 (%)	97.9 (96.6–98.0)	96.95 (95.7–98.0)	0.1[a]
Children with OSA (%)	26 (72.2%)	14 (77.7%)	0.7[b]
Oral breathing (%)	25 (69.4)	15 (83.0)	0.3[b]
Lip competence (%)	15 (41.6)	10 (55.0)	0.9[b]
Lip hypotonia (%)	27 (75.0)	13 (72.0)	0.8[b]
Normal tongue resting position (%)	5 (13.8)	2 (16.6)	0.7[b]
Normal tongue position during swallowing (%)	6 (16.6)	3 (16.6)	0.6[b]
Nasal cartilage hypotonia (%)	19 (52.7)	10 (55.5)	0.7[b]
Positive Glatzel test (%)	11 (30.5)	4 (22.2)	0.5[b]
Positive Rosenthal test (%)	14 (38.8)	6 (33.3)	0.7[b]

MT myofunctional therapy, *AHI* apnea–hypopnea index, *SpO_2* mean oxygen saturation, *OSA* obstructive sleep apnea

[a] Mann–Whitney *U* test

[b] Chi-square test

Table 2 Differences in IOPI measurements between the three groups of children

	MT group (36)	No-MT group (18)	Healthy group (38)	*p* values[a]
Tongue strength (kPa)	31.9 ± 10.7	32.4 ± 9.4	51.3 ± 13.6	Healthy vs no-MT group, <0.001
				Healthy vs MT group, <0.001
				MT group vs no-MT group, 0.5
Peak pressure (kPa)	34.2 ± 10.2	34.4 ± 9.9	54.2 ± 12.7	Healthy vs no-MT group, <0.001
				Healthy vs MT group, <0.001
				MT group vs no-MT group, 0.9
Endurance (s)	28.1 ± 8.9	23.3 ± 5.9	15.8 ± 7.2	Healthy vs no-MT group, <0.001
				Healthy vs MT group, <0.001
				MT group vs no-MT group, 0.06

MT myofunctional therapy

[a] Mann–Whitney *U* test

by age were previously validated by Potter et al. [19]. Maximal pressure for the tongue was determined by recording three maximal force efforts, each lasting approximately 1 s, with a 1-min rest period between trials.

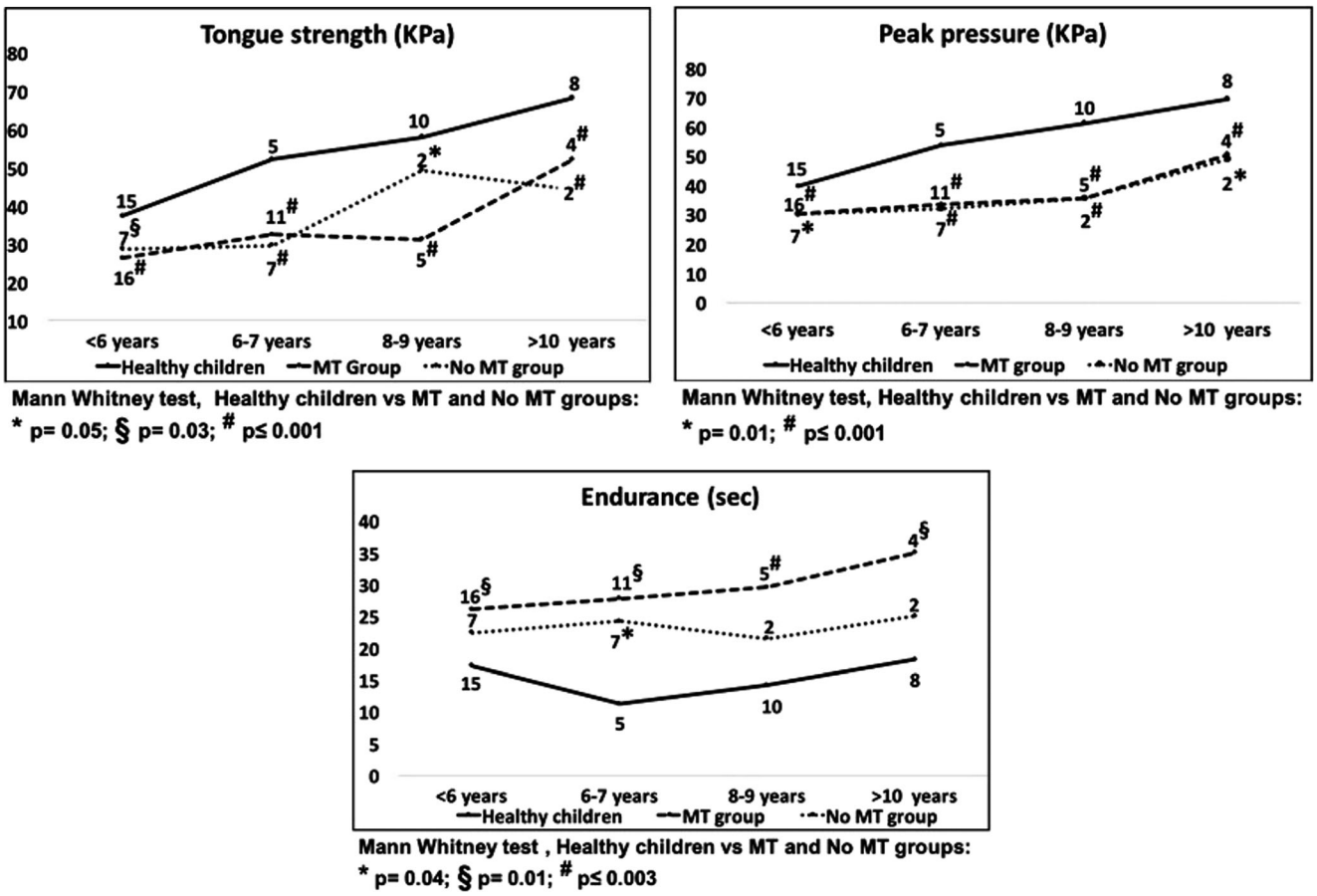

Fig. 1 Iowa Oral Performance Instrument (IOPI) measurements in the overall population divided according to age groups

Endurance was measured by asking the subjects to maintain 50% of their maximal pressure for as long as possible. The length of the endurance trial was measured in seconds.

Statistical analysis

All variables were tested for normality. Accordingly, the values are expressed as a number and percentage (%) for categorical variables, mean ± standard deviation (SD), or median and interquartile range (25–75 percentile) for continuous variables according to the normal distribution of the data. The chi-square test or Fisher's exact test was used for categorical variables, and the independent *t* test or Mann–Whitney test for continuous variables according to the normal distribution of the data. The Wilcoxon signed-rank test was used to compare paired data before and after MT. All the tests were two-tailed and a p value <0.05 was considered significant. The SPSS package (PASW Statistics for Windows, Version 23.0, Chicago: SPSS Inc. 2009) was used for all the analyses.

Results

We enrolled 54 children with sleep-disordered breathing (mean age 7.1 ± 2.5 years, 29 male) and 38 healthy children (7.8 ± 2.2, 25 male). According to the PSG records, 14 children suffered from primary snoring (PS, AHI 0.35 ± 0.3 ev/h) and 40 from mild–moderate OSA (AHI 2.2 ± 2.0 ev/h).

We divided the children into two groups: 36 were assigned to the MT group (mean age 6.7 ± 2.3 years, 14 male) and 18 (mean age 6.7 ± 2.8 years, 8 male) to the no-MT group. Table 1 shows the PSG parameters and myofunctional evaluation for each group.

SCR

No differences emerged between the SCR of the children in the MT group and those in the no-MT group (8.3 ± 2.1 vs 8.35 ± 2.1, $p = 0.9$), nor were any differences observed between the phenotype characteristics in the two groups: tonsillar hypertrophy (53 vs 77%), arched palate (75 vs 61%), skeletal malocclusions (44 vs 60%), and presence of obesity (2 vs 2%).

IOPI measurements

The IOPI, which was performed in all the SDB children and 38 healthy children, revealed significant differences between the MT and no-MT groups and the healthy children (Table 2). There were differences in IOPI measurements depending on age and between groups (Fig. 1). When we compared the IOPI measurements in the MT group before and after 2 months of treatment, we observed a significant improvement in all the parameters, whereas no differences were observed in the no-MT group (Fig. 2). Moreover, the myofunctional evaluation improved after 2 months of treatment in 18 children, as shown by a reduction in the oral breathing habit and lip hypotonia (Table 3).

When all the children with SDB repeated the pulse oximetry after 2 months, higher minimum and mean minimum oxygen saturation values and a lower oxygen desaturation index (ODI) were observed in the MT group, whereas no differences in the pulse oximetry values were observed in the no-MT group (Table 4).

Discussion

Our data demonstrate that MT reduced the respiratory symptoms during the night and oral breathing, and increased tongue tone as measured by means of the IOPI, an instrument easy to administer, in all the children with SDB. Moreover, subjects with SDB were found to have lower tongue strength as measured by the IOPI than healthy children.

The lower tongue strength and endurance observed in children with SDB are likely to be due to the persistence of oral breathing during sleep, which affects tongue position and strength as well as the orofacial muscles, and ultimately leads to abnormal craniofacial and airway development [8].

Oral breathing in children with SDB [20] results in an abnormal tongue position at rest and during sleep, which in turn reduces tongue movement and probably induces tongue hypotonia. These aspects are associated with skeletal malocclusions, as has been demonstrated by other authors [21], and

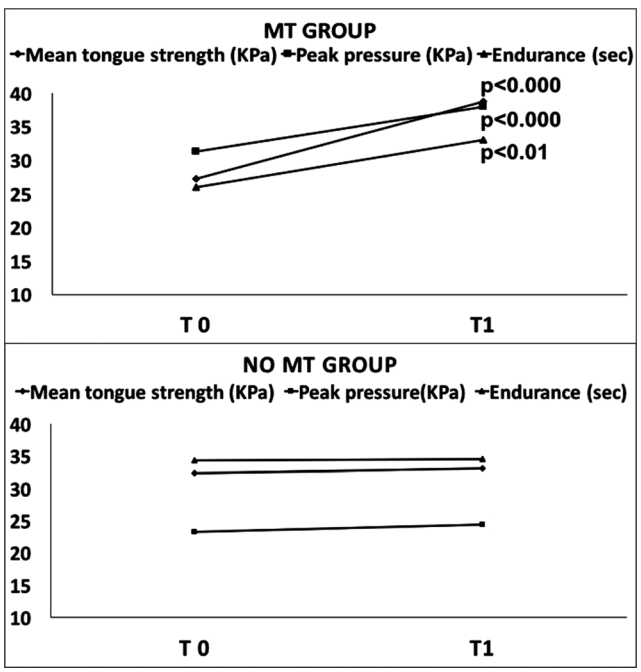

Fig. 2 Iowa Oral Performance Instrument (IOPI) measurements before and after 2 months of treatment in the MT and no-MT groups

Table 3 Myofunctional evaluation at T0 and at T1 in children with sleep-disordered breathing

	MT group (18)			No-MT group (18)		
	T0	T1	p^a	T0	T1	p^a
	18	18		18	18	
Oral breathing	15 (83.3)	3 (16.6)	0.0002	15 (83.0)	14 (78.0)	1.0
Lip incompetence	11 (61.1)	7 (39.0)	0.3	8 (44.4)	9 (50.0)	1.0
Lip hypotonia	14 (78)	6 (33.3)	0.003	13 (72.0)	12 (66.7)	1.0
Abnormal tongue resting position	17 (94.4)	12 (66.6)	0.03	16 (88.8)	16 (88.8)	1.0
Abnormal tongue position during swallowing	15 (83.3)	11 (61.1)	0.3	15 (83.3)	15 (83.3)	1.0
Nasal cartilage hypotonia	9 (50.0)	7 (38.8)	0.7	10 (55.5)	9 (50.0)	1.0
Positive Glatzel test	8 (44.0)	7 (38.8)	1.0	4 (22.2)	7 (38.8)	0.5
Positive Rosenthal test	8 (44.0)	4 (22.2)	0.3	6 (33.3)	8 (44.0)	0.7

MT myofunctional therapy

[a] Chi-square test

when the tongue is not in its normal position (i.e., lying against the maxillary incisors and hard palate) but sits on the mouth floor, the modelling role played by the tongue on the oral cavity, upon every deglutition, is strongly reduced [22].

Reduced tongue tone and endurance in children with SDB (i) may be a consequence of oral breathing and/or (ii) could be associated with sucking habits (bottle- or breastfeeding) and delayed chewing [23].

When the tongue is placed high in the palate, it correctly stimulates the intermaxillary synchondrosis, leading to normal orofacial growth and normal nasal breathing [24, 25]. Previous data have demonstrated that MT restores nasal breathing and re-establishes tongue position [10].

In our study, MT had a positive effect on tongue behaviour, restoring the normal tongue resting position after 2 months of treatment and enhancing tongue strength, maximum peak pressure, and endurance, as evaluated by means of the IOPI.

These aspects are associated with the restoration of nasal breathing and a reduction in mouth breathing, which in turn improve nocturnal breathing and reduce OSAS in both adult and pediatric populations [9, 10, 26].

These changes are related to nocturnal symptom reduction and result in a higher oxygen saturation, as has previously been demonstrated [10]. Moreover, the fact that oral breathing and lip hypotonia disappeared indicates that MT not only restores normal orofacial function but also may, if performed as a routine exercise, prevent tongue hypotonia [21]. The improvement in tongue function after 2 months of MT was accompanied by improved pulse oximetry values in mean oxygen saturation and a lower ODI. These results may be ascribed to the restoration of normal nasal breathing and the resulting increased supply of oxygen, nitric oxide, and alveolar ventilation during the night [27].

It is not clear how dental malocclusions are associated with stomathognatic myofunctional anomalies. There is, however, a consensus among orthodontists across Europe that myofunctional re-education of the oral–facial region plays an important role in correcting abnormal maxillary and mandibular growth, as well as in normalizing bite and teeth positioning. These effects are believed to be due to the restoration of normal local muscle activity [17].

The creation of oral–facial muscle re-education programs led to the creation of specific university training for the staff involved in such programs. Combined orthodontic and myofunctional re-education has since been applied to children with narrow jaws [8]. With regard to long-term outcomes, combined therapy has proved to be more effective than either treatment used on its own. Ever since maxillary–mandibular growth was shown to be

Table 4 Oxygen saturation values before and after myofunctional treatment

	MT group (18)			No-MT group (18)		
	T0	T1	p^a	T0	T1	p^a
Oxygen desaturation index (ODI)	5.9 ± 2.3	3.6 ± 1.8	0.000	6.3 ± 2.7	7.1 ± 3.2	0.7
Mean oxygen saturation (%)	96.4 ± 0.6	97.4 ± 0.7	0.000	96.1 ± 2.2	96.2 ± 1.5	0.9
Minimum oxygen saturation (%)	91.1 ± 1.4	91.2 ± 1.3	0.9	85.2 ± 4.1	87.7 ± 4.9	0.2
Mean minimum oxygen saturation (%)	94.3 ± 1.3	95.4 ± 1.3	0.000	93.7 ± 2.1	94.2 ± 1.4	0.5

[a] Wilcoxon signed-rank test

involved in SDB, children have been treated with both myofunctional re-education and orthodontia [8, 17].

We know that tongue function may play a role in stomatognathic remodelling and that the restoration of correct normal tongue function needs to be integrated in orthodontic and surgical treatments.

Limitations of the study

One limitation of this study is that the polysomnography was not repeated in children with SDB after 2 months of MT. However, the pulse oximetry, which was repeated, yielded good results. The decision not to repeat the polysomnography was taken to avoid the high costs involved in readmitting the children to hospital, to spare the children and their families any further discomfort as well as because of the parents' opposition, who were already satisfied with the improvement in their children's respiratory symptoms and deemed a repeat examination unnecessary.

Moreover, this study included children with mild–moderate OSA; further research is needed to determine if MT could be effective in patients with different OSA severities and complementary to other treatments.

Conclusions

Children with SDB may exhibit tongue thrusting and abnormal swallowing patterns caused by nasal obstruction and a persistent mouth breathing posture. MT may be used to integrate medical and surgical treatments for OSA and help to restore a normal resting posture of the tongue; appropriate oral, lingual, and facial muscle patterns; nasal breathing; normal lip posture; and a correct swallowing pattern.

Compliance with ethical standards

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Conflict of interest The authors declare that they have no conflict of interest. All authors certify that they have no affiliations with or involvement in any organization or entity with any financial or non-financial interest in the subject matter or materials discussed in this manuscript.

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All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Additional informed consent was obtained from all individual participants for whom identifying information is included in this article.

This article does not contain any studies with animals performed by any of the authors.

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Declaration of authorship

Prof. Maria Pia Villa had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.