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ORIGINAL ARTICLE

Passive myofunctional therapy applied on children with obstructive sleep apnea: A 6-month follow-up

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KEYWORDS
children;
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Background/purpose: Myofunctional therapy is one of the recommended treatments for obstructive sleep apnea, but the level of compliance has often been low in children. This study aims to investigate the therapeutic effect of passive myofunctional therapy using an oral appliance during sleep in children suffering from obstructive sleep apnea.
Methods: Twenty-nine children who suffered from obstructive sleep apnea were divided into two groups: premature children and full-term children. All children wore an oral device to induce their tongue muscle activity during sleep for 6 months. Polysomnography during sleep was performed before and 1 week after the end of 6-month treatment.
Results: Both groups showed positive polysomnographic changes. Full-term children had a significant decrease in the apnea–hypopnea index, hypopnea index, and percentage of arousals. Prematurely born children had a significant decrease in the apnea–hypopnea index during rapid eye movement sleep and in the mean heart rate during sleep.
Conclusion: Using a specialized oral device to perform myofunctional therapy during sleep may improve the breathing during sleep of children with obstructive sleep apnea.

Conflicts of interest: The authors have no conflicts of interest relevant to this article. Dr Hervy is the owner of the Myonix device patent.

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Introduction

A high and narrow hard palate and obstructive sleep apnea (OSA) and/or hypopnea are found in a very high percentage (>80%) of children with premature birth.[1–4] These children also have a high prevalence of malocclusion and often need orthodontic treatment.[5] Although most premature infants will catch up with their peers developmentally later on (particularly during adolescence),[6–8] they often have a lower weight, shorter height, and smaller head circumference by the age of 10 years, compared with full-term children.[6–9] They also tend to have a shorter anterior cranial base, less convex skeletal profile, and shorter maxillary length.[9]

Villa et al[10] studied a mandibular advancement device (MAD) for the treatment of OSA in children and reported that wearing a MAD for 6 months could reduce the apnea–hypopnea index (AHI) from 7.1 to 2.6 and improve the clinical respiratory symptom. Myofunctional therapy (MFT) has been administered to adults and children with OSA to improve their breathing during sleep, and it has decreased the AHI by approximately 50% in adults and 62% in children.[11,12] The therapy aims to improve the tongue muscle, particularly in hypotonic premature infants.[1] However, the level of compliance for MFT has often been low in children. Economic and social conditions could also impact the performance of daily oral–facial exercises. The oral appliance studied in the current research is designed by one of the authors to help children perform oral exercises at bedtime and during sleep. Figure 1 shows the device with a bead placed close to the tip of the tongue. Moving along an axis, the bead is supported by a light frame mounted on the lower teeth, similar to other oral appliances. The device is designed on the basis that intrusion of a foreign object close to the tip of the tongue stimulates tongue activity at least during light stages of sleep. Better tongue functioning means better overall tongue position at rest, which is important for normal oral cavity growth and maxillary arch expansion. Proper positioning of the tongue through MFT has been demonstrated to improve nasal breathing, mandibular growth, and facial appearance.[13]

This report presents the results of an objective study using polysomnography (PSG) to evaluate the short-term effect of passive MFT during nocturnal sleep by inserting an oral appliance in the mouth of children with OSA, including those who were prematurely born.

Methods

Patient recruitment and procedures

The study protocol was approved by the Institutional Review Broad of the Human Investigation Committee of the Chang Gung Memorial Hospital. Informed consent from the legal guardian of each participant was obtained prior to investigation.

As shown in Table 1, 29 patients (23 boys and 6 girls; mean age: 9.76 ± 3.54 years; range: 3–15 years) with a diagnosis of pediatric OSA based on the International Classification of Sleep Disorders-Third Edition participated in the study. The inclusion criteria for this study, based on the results of PSG, were as follows: (1) AHI ≥ 1 event/h and (2) Respiratory Disturbance Index ≥ 5 events/h. The following demographic and clinical information of all participants was collected during their initial visit to the sleep laboratory: age, sex, body mass index (BMI), body weight and height, gestational age, and birth body weight. All children had been diagnosed with OSA based on their clinical complaints/symptoms and the results of PSG at the Sleep Center of the Chang Gung Memorial Hospital. Children were divided into two groups, based on their gestational age: "premature" (<37 weeks) and "full term". The exclusion criteria for the study were as follows: epilepsy, head injury, severe developmental delay and mental retardation, autism, schizophrenia, severe depression, or inability to cooperate for the measurement of PSG or for the fabrication of oral devices.

Pediatric OSA is different from the adult type as most cases of pediatric OSA are mild to moderate. Most of the participants had undergone adenotonsillectomy before the study and had a residual AHI when the study began. The rest did not have adenotonsillar hypertrophy, so they could be included in the study.

All participants underwent PSG before treatment with the oral appliance and after 6 months of wearing the device nightly.

PSG during sleep

A Compumedics sleep system was used to monitor the following: electroencephalography (C4/A1, C3/A2, Fp1/T3, T3/O1, Fp2/T4, T4/O2, Fp1/C3, Fp2/C4), electrooculogram, chin and leg electromyography, electrocardiography with a modified V2 lead, body-position sensor, nasal cannula/pressure transducer, mouth thermistor, thoracoabdominal and plethysmographic bands, neck microphone, and finger pulse oximetry. Scoring was performed by an individual who was not involved in the study and was blind to the settings (i.e., pre- or post-treatment).

Oral appliance

The appliance is a one-piece, custom-made adjustable oral device for advancing the mandible. A bead is mounted on the lower part of the frame for the tip of the tongue to roll, which in turn places the tongue in a forward position so as to open the airway (Figure 1). The amount of mandibular advancement associated with the wearing of the device was 50% of the maximum mandibular advancement. Patients were instructed to wear their appliances and use their tongue to roll the bead (i.e., passive MFT) during sleep every night. Parents kept sleep logs to record the nightly wear by all children for 6 months. The study participants did not receive active MFT.

Statistical analysis

Data were analyzed using a statistical software package (SPSS Inc. Released 2009. PASW Statistics for Windows, Version 18.0. Chicago: SPSS Inc.). Descriptive statistics were presented as means and standard deviations.

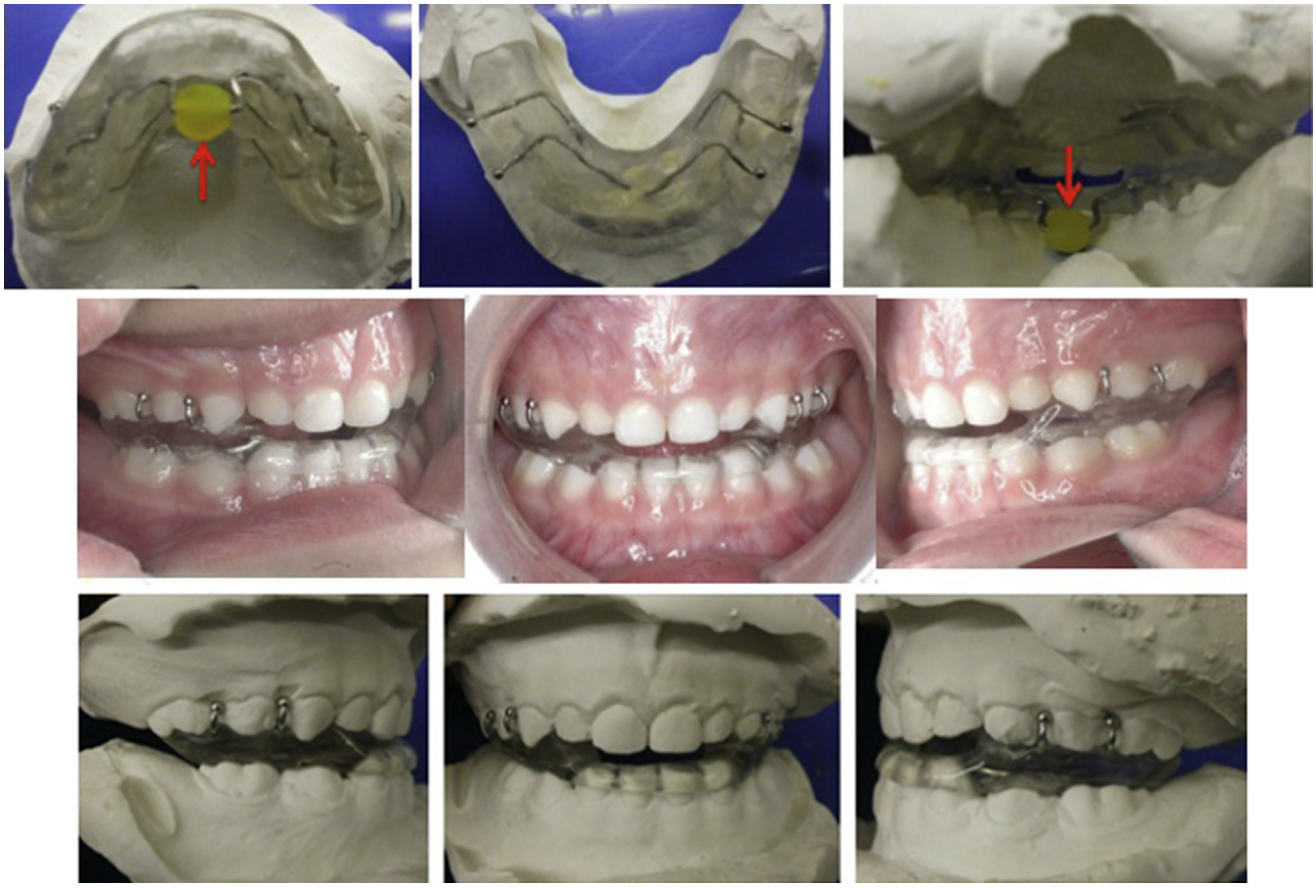

Figure 1 The oral appliance is a one-piece, custom-made adjustable oral device for advancing the mandible. A bead (red arrow) is mounted on the lower part of the frame for the tip of the tongue to roll, which in turn places the tongue in a forward position so as to open the airway. This appliance is designed by Dr Michèle Hervy-Auboiron.

Chi-square test was used to test whether there were gender differences between full-term and preterm groups. Mann—Whitney *U* test was used to test whether there were significant differences in basic clinical data at the baseline. Wilcoxon signed-rank test was used to test whether there were significant differences in the PSG data before and after treatment with the oral device. The level of significance was set at $p < 0.05$.

Results

Twenty-nine children were enrolled in this study. The data in Table 1 show no significant differences in sex, age, and body weight distributions between the two groups (premature and full-term birth) at the time of recording.

According to the questionnaires completed, parents were relieved to not have to perform daily active

Table 1 Demographics of the participants.

	Full term ($N = 18$)	Premature ($N = 11$)	Total ($N = 29$)	*p*
Sex, *n*				
Male	16 (88.9%)	7 (63.6%)	23 (79.3%)	
Female	2 (11.1%)	4 (36.4%)	6 (20.7%)	0.103[a]
Age (y)	10.44 ± 3.18	8.64 ± 3.96	9.76 ± 3.54	0.187[b]
Gestational age (wk)	39.6 ± 0.3	33.2 ± 1.2	36.8 ± 4.3	<0.001[b]
Birth body weight (g)	3491.8 ± 157.9	2104.0 ± 256.0	2881.2 ± 991.8	<0.001[b]
Body weight (kg)	38.2 ± 19.4	24.8 ± 15.8	33.1 ± 19.0	0.053[b]
Body height (cm)	134.5 ± 19.5	117.9 ± 20.0	128.2 ± 20.9	0.036[b]

All data are listed as means and standard deviations.
[a] Chi-square test.
[b] Mann—Whitney test.

Table 2 Polysomnography data before and after treatment.

	Intragroup comparison									Intergroup comparison	
	Full term ($N = 18$)			Premature ($N = 11$)			Total ($N = 29$)			Baseline	After treatment
	Baseline	After	p	Baseline	After	p	Baseline	After	p	p	p
Body weight	38.2 ± 19.4	43.5 ± 17.3	0.006**	24.8 ± 15.8	29.7 ± 16.1	0.160	33.1 ± 19.0	37.7 ± 17.7	0.004**	0.053	0.007**
Body height	134.5 ± 19.5	146.0 ± 19.6	0.003**	117.9 ± 20.0	130.2 ± 21.8	0.012*	128.2 ± 20.9	139.4 ± 21.5	0.001**	0.036*	0.014*
BMI	19.7 ± 5.3	19.5 ± 3.2	0.247	15.77 ± 5.07	16.35 ± 3.81	0.575	18.2 ± 5.4	18.2 ± 3.7	0.763	0.023*	0.004**
AHI in sleep	4.9 ± 4.9	2.1 ± 2.9	0.041*	6.0 ± 7.6	1.8 ± 1.9	0.069	5.4 ± 5.9	1.9 ± 2.5	0.005**	>0.99	0.856
AHI in REM	10.1 ± 11.8	6.0 ± 12.2	0.169	10.6 ± 12.4	5.9 ± 10.9	0.036*	10.3 ± 11.8	5.9 ± 11.4	0.020*	0.856	0.897
AI = (AHI − HI)	0.77 ± 1.1	0.74 ± 1.36	0.507	1.1 ± 1.3	0.65 ± 0.7	0.352	0.9 ± 1.2	0.7 ± 1.1	0.201	0.484	0.737
HI = (AHI − AI)	4.19 ± 4.8	1.33 ± 2.4	0.029*	2.70 ± 2.7	1.15 ± 1.6	0.128	3.63 ± 4.1	1.25 ± 2.02	0.007**	0.336	0.698
Desaturation index	3.73 ± 4.9	2.15 ± 3.3	0.444	3.23 ± 3.3	2.29 ± 2.3	0.263	3.54 ± 4.3	2.21 ± 2.8	0.267	0.816	0.776
Efficiency %	82.7 ± 14.3	89.1 ± 8.6	0.091	84.7 ± 10.4	85.8 ± 5.5	0.674	83.4 ± 12.8	87.7 ± 7.4	0.121	0.517	0.286
Awake %	12.70 ± 14.4	6.35 ± 5.6	0.021*	12.47 ± 10.5	8.80 ± 5.8	0.208	12.61 ± 12.9	7.38 ± 5.6	0.009**	0.517	0.698
REM %	18.8 ± 4.7	16.1 ± 5.0	0.424	22.7 ± 4.5	18.23 ± 8.0	0.401	20.28 ± 4.9	17.0 ± 6.33	0.212	0.003**	0.363
Stage N1%	9.59 ± 3.5	8.86 ± 3.4	0.625	14.36 ± 12.6	11.50 ± 6.5	0.401	11.40 ± 8.3	9.97 ± 4.9	0.286	0.856	0.698
Stage N2%	42.72 ± 7.4	46.92 ± 6.5	0.285	33.79 ± 6.8	42.40 ± 10.5	0.263	39.33 ± 8.3	45.02 ± 8.5	0.126	0.036*	0.014*
Stage N3%	28.74 ± 7.2	28.08 ± 6.1	0.859	29.13 ± 13.2	27.89 ± 9.2	0.400	28.89 ± 9.7	28.00 ± 7.3	0.747	0.698	0.220
Total sleep time	366.1 ± 81.2	398.2 ± 32.5	0.131	391.7 ± 44.5	397.8 ± 22.9	0.779	375.8 ± 69.7	398.0 ± 28.1	0.165	0.087	0.776
Sleep latency	16.44 ± 13.3	19.00 ± 25.7	0.894	11.82 ± 8.2	23.44 ± 12.8	0.069	14.69 ± 11.7	20.87 ± 20.9	0.184	0.776	0.003**
Arousal index	11.25 ± 5.4	10.26 ± 4.3	0.328	11.05 ± 6.1	10.534 ± 6.5	0.484	11.18 ± 5.6	10.38 ± 5.2	0.227	0.509	0.517
Mean heart rate	75.93 ± 11.8	68.79 ± 8.2	0.109	80.45 ± 10.7	71.25 ± 9.6	0.030*	77.65 ± 11.4	69.83 ± 8.6	0.007**	0.241	0.201
Snore index	155.1 ± 215.9	71.6 ± 136.1	0.248	140.1 ± 242.0	46.76 ± 84.5	0.208	149.4 ± 221.9	61.1 ± 115.0	0.059	0.897	0.737
Mean SpO_2 %	97.47 ± 0.7	97.70 ± 0.46	0.590	96.73 ± 1.35	97.38 ± 0.92	0.458	97.19 ± 1.06	97.56 ± 0.69	0.375	0.286	0.660

Intragroup comparison: Wilcoxon signed-rank test; intergroup comparison: Mann–Whitney test.

* $p < 0.05$.

** $p < 0.01$.

AHI = apnea–hypopnea index; AI = apnea index; BMI = body mass index; HI = hypopnea index; REM = rapid eye movement; SpO_2 = oxyhemoglobin saturation by pulse oximetry.

myofunctional exercises with the children and that children accepted nightly use of the device without complaint or objection. No side effect was reported by children or parents.

The PSG results before and after treatment with the oral device are shown in Table 2, including both intra- and intergroup comparisons. After the treatment, the full-term group had a significant increase in body weight and body height as well as a significant decrease in the AHI in sleep, hypopnea index (HI), and percentage of wake time. The premature group had a significant increase in body height and decrease in the AHI related to rapid eye movement (REM) sleep and the mean heart rate. For both groups as a whole, there was a significant increase in body weight and height, and a significant decrease in the AHI, AHI related to REM sleep, HI, percentage of wake after sleep onset, and mean heart rate.

The intergroup comparison shows a significantly lower body height, BMI, and percentage of Stage 2 sleep in the preterm group at the baseline, although the percentage of REM is higher in the preterm group. After 6 months of treatment with the oral device, significantly lower body weight, body height, BMI, and percentage of Stage 2 sleep are seen in the preterm group, while sleep latency is higher in the preterm group.

Discussion

This is the first investigation of a device designed to stimulate tongue activities at least during light stages of sleep, in order to obtain similar results to active MFT. Active MFT requires the involvement of at least one parent and the child for a minimum of 10 minutes of active exercises both in the morning and in the evening. Maintaining this routine has proved challenging with young children and compliance has been limited at best, particularly if there is no regular contact with an MFT specialist who can provide support to the parent and the child.

Pediatric OSA is different from adult OSA, as most pediatric OSA is mild (AHI = 1—5) to moderate (AHI = 5—10) and rarely severe (AHI > 10). Moreover, adults with moderate to severe OSA will need continuous positive airway pressure or maxillomandibular advancement treatment. Soft tissue surgery or dental treatment could decrease the severity of OSA but would not cure the OSA problem. For children with moderate to severe OSA, continuous positive airway pressure or adenotonsillectomy is the recommended treatment. Therefore, considering the risks associated with moderate to severe OSA and clinical ethics when designing the study, we only recruited children with mild to moderate OSA. Furthermore, because the brain physiology of children is different from that of adults, hypopnea and respiratory effort-related arousals (<3% hypopnea) appear more prevalent in pediatric OSA, but not apnea. Thus, more improvement was seen in the HI, but not in the apnea index.

In Taiwan, there are relatively few patients with pediatric OSA (under 12 years of age) who are also obese.[14] Moreover, obese pediatric patients with OSA usually suffer from the severe type of OSA, who, thus, would not be suitable candidates for passive MFT using this oral appliance.

As the intergroup comparison shows, the preterm group has significantly lower weight, height, BMI, and growth rate than their full-term peers, mirroring findings of previous studies.[6—8] Preterm children often have a higher percentage of REM (as shown at the baseline), but, after the treatment, the percentage of REM decreased in both groups. After the treatment, sleep latency also increased in both groups, especially in the preterm group. This may be attributable to the smaller mouth and higher sensitivity to oral appliances of preterm children.

Meanwhile, there are some limitations to our study. First, the sample size of the preterm group is small. Hence, the explanatory power of the study will be limited, although our study showed the feasibility of using the device and, despite the small sample size, very clear and significant improvement post-treatment.

Second, the study period is relatively short. According to the study of Guimaraes,[15] MFT for as short as 3 months could still yield a significant improvement in adult OSA patients. Despite only 6 months of wearing the oral appliance nightly by the participants in our study, we saw significant improvements such as decreases in the AHI, AHI related to REM sleep, HI, percentage of wake after sleep onset, and mean heart rate. We will continue this cohort study to observe the long-term effect of passive MFT using this oral appliance as well as whether the therapeutic effect remains after the use of the oral appliance is stopped.

Third, there is no control group in the study, for example, a group without using the oral device or a group with active MFT.[11,12] This oral device is composed of a tongue training target set on mandibular advancement. Efforts were made to rule out the effect of the usual MAD, i.e., a device without the rolling bead. The amount of mandibular advancement associated with the wearing of the device was 50% of the maximum mandibular advancement. Despite these efforts, it remains possible that some of the positive gains could partially be related to the effect of a plain MAD. With the current product design, a control group using MADs without the tongue bead will be needed to further delineate the contributors to these positive gains.[10]

Our results, however, showed that the usage of such an oral device only during the nocturnal period had a beneficial effect, as documented by the PSG data. Spontaneously children with a small oral cavity associated with maxilla—mandibular involvement do not improve particularly in such a short time.

In conclusion, a device for passive MFT has shown no negative effect after 6 months of usage, while improving sleep and respiratory variables during sleep. Our study provided preliminary evidence to support follow-on larger studies with more adequate controls.

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