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Original Article

Neutral supporting mandibular advancement device with tongue bead for passive myofunctional therapy: a long term follow-up study

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ABSTRACT

Backgrounds: Myofunctional therapy has been reported to be a valid adjunct treatment to OSA, but compliance was mentioned as an issue. We performed a prospective study on age matched randomized children submitted to myofunctional therapy (MFT) or to a functional device used during sleep (passive MFT).

Methods: 110 children 4 to 16 were recruited for the study, 54 children were in the MFT group [A] while 56 were in the "nocturnal device" group [B]. Clinical evaluation, polysomnography and cephalometric X-Rays were performed at baseline, 6 months and 12 months, with clinical follow-up at 3 months.

Results: MFT group show very important absence of compliance, at six months only 23 subjects participated and only 10/23 had been compliant with treatment. None came back for research investigation at 12 months. 48/56 of passive MFT children ended the research protocol at 12 months. Comparison of baseline to 6 and 12 months data showed that all children with passive MFT improved (PSG and cephalometrics) and had nasal breathing during sleep at 1 year, and no negative effect of device were noted. The 10 children compliant with MFT showed clear improvement of sleep related breathing with also changes at cephalometric −X−rays.

Conclusion: Compliance is a major problem of MFT, and MFT will have to take into consideration the absolute need to have continuous parental involvement in the procedure. Passive MFT gives many more positive results, but potential negative effects of device on other jaw will have to be continuously evaluated.

1. Introduction

Sleep-disordered breathing (SDB) is a common health problem within the pediatric population [1]. In 1981, Guilleminault et al. published a review of fifty pediatric patients [2,3] which demonstrated that the clinical features of pediatric OSA were different from adults. Among the types of pediatric SDB, obstructive sleep apnea (OSA) has the highest prevalence, but its pathophysiology continues to be unclear. The underlying causes of pediatric OSA are complex. Such as adenotonsillar hypertrophy, obesity, anatomical and neuromuscular factors, and hypotonic neuromuscular diseases are also involved. Currently, the main cause of pediatric OSA is most commonly hypothesized to be a result of an anatomically or functionally narrowed upper airway. Related research in recent years showed that anatomic features of children with craniofacial anomalies are highly related to pediatric OSA, as shown in our review of 1000 children polysomnographically monitored for suspicion of OSA with children with obesity eliminated from the survey, and after eliminating children with very large tonsils (n = 114), 92.88% presented with abnormalities of the orofacial complex.as shown by clinical evaluation by both sleep-specialist and orthodontist [4,5] Therefore, the established first-line treatment,

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adenotonsillectomy (AT), is unable to eradicate pediatric OSA completely, and high levels of relapse continues to be a point of contention [6]. The premature infants show very high rates of pediatric OSA [7]. It means the initial oral facial anatomic build-up of an individual as a clear impact on the risk of collapsibility of the UA during sleep [4,7]. As there is a continuous interaction between orofacial functions and oro-facial growth after birth. The dysfunctions identified to date impact orofacial development leading to sleep-disordered-breathing through changes in the orofacial growth [4,5,7,8]. Recently, myofunctional therapy (MFT) has been used in different parts of the world [9,10] for many years and it has been suggested as an adjunct treatment of SDB in children [4,9–14]. Results of studies done on children with orthodontic problems have shown that isolated extensive and well-controlled MFT can lead to return to normal orofacial anatomy [11–14]. In adults, there is reported improvement of OSA and snoring also [9,10,16]. But the use of MFT alone when dealing with pediatric SDB has not been widely investigated and the long-term effects of MFT on SDB are still unknown. Problems with MFT have been reported [1]: current forms of MFT are difficult for children younger than 4-years of age [2], the poor compliance with daily exercises and the absence of continuous parental involvement with the training exercises of the child are major causes of failure of treatment. Preliminary studies indicated that usage of a functional oral appliance thought to induce some extra muscle activity while asleep (also called “passive myofunctional therapy”, PMFT) may decrease mouth breathing and may have an impact on the position of the mandible [16]. Such changes may lead to improvement of the narrow upper-airway during sleep, and may improve the snoring and pediatric OSA. Our study explored the effectiveness of MFT and PMFT using a previously described dental device on pediatric OSA and the long-term effects of these approaches on OSA and craniofacial growth.

2. Methods

2.1. The study had the following design

2.1.1. Patients recruitment

(1) Children between 4 and 16 years of age, diagnosed with OSA based on clinical evaluation and nocturnal—in laboratory-polysomnography-PSG-with an apnea-hypopnea-index (AHI) > 1 even/hr or a respiratory-disturbance-index (RDI) > 5 even/hr; and
(2) + Either children previously diagnosed with pediatric OSA who had undergone adenotonsillectomy, but with residual AHI > 1 even/hr or RDI > 5 even/hr at six months post T&A. + Or children diagnosed with pediatric OSA but without evidence of adenotonsil hypertrophy post-ear-nose and throat-ENT-evaluation.
(3) Recruited children were age matched and were randomized to be treated (a) either with MFT (total 20 min/per day) for 1 year (b) or treated with a previously described and specifically design oral appliance with a tongue bead [15] during sleep (passive MFT) for 1 year. 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.

2.1.2. Procedures

Evaluation of the 2 groups was planned to be at baseline and subsequently at 3-, 6-, and 12-months after MFT or PMFT treatment, with clinical evaluation, PSG and lateral cephalometrics evaluating bone structure development at baseline, 6 and 12 months.

2.1.3. PSG during sleep

A Neurovirtual BWIII PSG Plus sleep system™ (Fort- Lauderdale. Fl. USA) was used. The following variables were recorded: electroencephalography (F3-M2, F4-M1, C3-M2, C4-M1, O1-M2, O2-M1), electro-oculogram (EOG), chin and leg electromyography (EMG), electrocardiography (ECG) with a modified V2 lead, body-position sensor, nasal cannula/pressure transducer, mouth thermistor, thoracic and abdominal plethysmography bands, neck microphone, and finger pulse oximetry.

Scoring was performed by an individual not involved in the study and blind to all conditions. The American Academy of Sleep Medicine “recommended” criteria were used to score PSG.

2.1.4. MFT

MFT is comprised of isotonic and isometric exercises that target oral (lip, tongue) and oropharyngeal structures (soft palate, lateral pharyngeal wall). MFT [10,14,15] aims to obtain appropriate head posture and positioning of the tongue on the palate against the upper teeth, appropriate swallowing and mastication using both sides and posterior chewing, appropriate breathing through the nose while keeping the mouth closed, and appropriate speech and articulation. MFT requires active parental involvement to obtain good results. Parents and children had initial training with a specialist and parents were asked to supervise a minimum of 20 min of exercise daily. The MFT specialist was available to parents and regular clinical follow-up were scheduled. This regular daily activity was called “active MFT”.

2.1.5. Oral appliance

The ‘passive MFT’ short term used was reported recently [16]. The amount of mandibular advancement associated with the wearing of the device at rest-supine awake was 50% of the maximum mandibular advancement (Fig. 1, patent number: EP 13753289.1; US14/420499). The device aims at raising the tongue position during sleep and the mandibular part is more a support of the bead located high near the hard palate than aimed at a therapeutic goal. This part is not advanced overtime. Patients were instructed to wear their appliance nightly, the appliance been placed just at bed-time and to use their tongue to roll the bead (ie, passive MFT) just before falling asleep. Parents kept sleep logs to record the nightly wear by all children for 12 months. These sleep logs were used to calculate compliance. The device is inspected on a monthly basis by the orthodontist and parents have a direct phone number to contact if any problem or change is noted in the device.

2.1.6. Cephalometrics evaluating

Fig. 2 showed the Craniofacial skeletal landmarks and airway landmarks.

3. Statistical analysis

General descriptive statistics, ANOVA, Wilcoxon signed rank test and Mann–Whitney analyses of the treatment results from the lateral cephalometric X-ray, and PSG reports were performed using SPSS-18.

4. Results

Total 110 pediatric OSA were enrolled. Fifty-four children were randomized to receive MFT (Group A). But only 23 children completed the six months PSG study. Out of these 23, only 10 children had good compliance (compliance rate >80%) and completed pre and post cephalometric test. At 1 year follow-up almost all patients had stopped performing MFT and none wanted to participate to the study. Group B enrolled 56 pediatric OSA who received oral

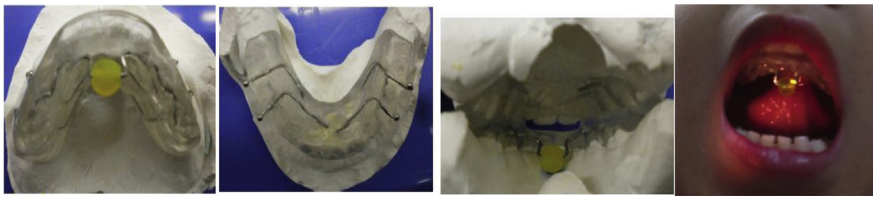

Fig. 1. The oral appliance in the present study is combined with MAD and tongue bead designed by Dr. Michelle Hervey (patent number: EP 13753289.1; US14/420499).

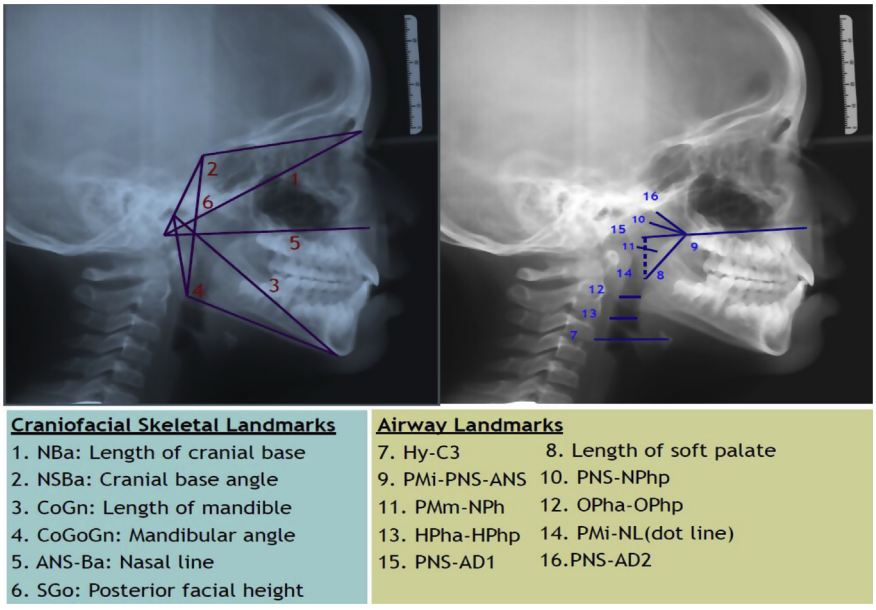

Fig. 2. Cephalometric landmarks.

device (PMFT) treatment and 52 children completed 0.5 year research. Only 48 children with good compliance (>80%) and could complete a year of research and received PSG and cephalometric test. The data showed no significant differences in age (group A:7.02 ± 2.44; group B:7.97 ± 3.08, p = 0.338), sex (50% boy; 64.6% boy; p = 0.37), and BMI (15.61 ± 1.74; 17.04 ± 3.05, p = 0.080) distributions between the two groups.

According to the questionnaires administered with dental device, children accepted nightly use of the device without complaint or objection. No side effect was reported by children or parents.

Considering the very important "drop-out" rate of subjects in group A, we firstly performed analyses on Group A subjects. We considered each subject as its own control and compared baseline to post treatment. PSG results are presented in Table 1. At six months follow-up, significant differences were already noted involving the respiratory disturbance index (RDI) (from 3.53 ± 2.01 to 2.57 ± 1.76 events/hour, p = 0.032), respiratory effort related arousal (RERA) (from 5.87 ± 8.96 to .1.74±2.78 events/hour, p = 0.048) and sleep latency (from 16.20 ± 12.83 to 10.78 ± 9.49 min, p = 0.036) that showed significant improvement after 0.5 year of treatment. But none of the subjects in group A came back for complete evaluation at 12 months.

Table 2 is the PSG outcome of 0.5 year and one year of treatment with PMFT. Many significant findings were noted. Significant increase in body weight (p < 0.001) and body height (p < 0.001) with age. Normal increase for age in BMI was noted as well as significant decrease of the AHI during total sleep (from 6.00 ± 7.23 to 2.64 ± 2.47 and 2.44 ± 2.28 events/hour, p = 0.001) and during REM sleep (p = 0.023), significant decrease in the hypopnea index (HI) (p = 0.001), the snoring index (p = 0.046), the sleep efficiency (p = 0.044) and the hypopnea count (p = 0.006). Respiratory events measured without usage of oral device at time of PSG recording, showed clear improvement without any other treatment approach than "passive −MFT" for one year.

Despite the very large drop-out at 1 year, we performed an " intergroup comparison" at 6 months when a sufficient number of subjects (n = 23) reported having performed MFT-even if we limited compliance. Table 3 showed results: AHI in sleep (p < 0.001), HI(p = 0.001), hypopnea count (p = 0.012), and percentage of awake (p < 0.001) had more improvement in group B (oral device) than group A (MFT). But sleep latency (p = 0.009) was significantly increased in group B (PMFT) than group A. It may be due to close mouth breathing under treatment with oral device.

We considered craniofacial development and airway change after treatment with MFT and oral device (PMFT) by cephalometric test. We compared only the children with good compliances for MFT ie n = 10 and the subjects that used the device regularly ie n = 48. This comparison could be done only at the 6 months follow-up as none of the MFT subjects came back for complete evaluation at 12 months follow-up. As shown in Table 4, subjects with oral device had an A point-nasion-B point (ANB) angle in normal range after wearing oral device for 0.5 year, strongly suggesting absence of significant side-effect of wearing the device on a nightly basis. The investigation indicated that the oral device significantly improved the width of the airway at the level of nasopharynx (min-RGA (Minimal retroglossal airway), PNS-AD2 (Distance from PNS to the nearest adenoid tissue measured along the line perpendicular to S-BA)). Interestingly the patients who received MFT and who had

Table 1
PSG results of treatment with MFT.

	PSG Pre(n = 23) Mean ± SD	PSG-0.5 y (n = 23) Mean ± SD	PSG-1y (n = 0)	p-value
Body Hight (cm)	106.35 ± 13.40	115.60 ± 12.79	–	<0.001***
BMI	15.61 ± 1.74	15.76 ± 2.12	–	0.589
AHI in sleep (event/hr)	2.47 ± 1.31	2.26 ± 1.84	–	0.492
AHI in REM(event/hr)	5.27 ± 4.15	4.99 ± 6.18	–	0.799
AI (event/hr)	1.43 ± 1.04	1.03 ± 02	–	0.082
HI (event/hr)	1.05 ± 0.92	1.23 ± 1.25	–	0.518
Desaturation Index	1.64 ± 1.12	1.71 ± 2.22	–	0.877
RDI	3.53 ± 2.01	2.57 ± 1.76	–	0.032*
Efficiency %	89.37 ± 5.01	90.41 ± 78.32	–	0.312
Awake %	6.67 ± 4.58	7.04 ± 5.17	–	0.759
REM %	24.31 ± 4.66	20.13 ± 7.0	–	0.022*
Stage 1%	8.12 ± 4.43	12.15 ± 6.83	–	0.041*
Stage 2%	38.82 ± 7.71	39.47 ± 9.87	–	0.736
Stage 3%	28.75 ± 6.63	27.38 ± 7.53	–	0.485
Total sleep time (mins)	394.43 ± 22.73	398.33 ± 21.72	–	0.504
Awake Count	12.70 ± 4.36	14.35 ± 5.52	–	0.170
Sleep Latency (mins)	16.20 ± 12.83	10.78 ± 9.49	–	0.036*
REM Latency (mins)	64.5 ± 27.16	94.5 ± 56.34	–	0.023*
Arousal Count	43.83 ± 14.27	40.52 ± 11.15	–	0.321
Arousal Index	7.96 ± 2.74	7.2 ± 1.97	–	0.219
Mean Heart Rate	82.14 ± 14.04	78.23 ± 10.67	–	0.098
PLM Index (event/hr)	0.30 ± 1.00	1.14 ± 2.66	–	0.124
Snore Index (event/hr)	136.32 ± 144.80	122.38 ± 123.58	–	0.683
Obstructive Apnea	0.39 ± 0.78	0.48 ± 0.99	–	0.648
Central Apnea	6.87 ± 5.50	4.91 ± 5.53	–	0.161
Mixed Apnea	0.74 ± 1.86	0.48 ± 0.85	–	0.503
Hyponea	5.87 ± 5.11	6.91 ± 7.17	–	0.515
RERA	5.87 ± 8.96	1.74 ± 2.78	–	0.048*
Mean SaO2%	97.74 ± 0.62	97.83 ± 0.39	–	0.539

t-test was performed to analyze the 0.5 year of treatment results.
* P<0.05, ***p<0.001.
AHI, apnea-hypopnea index; AI, apnea index; HI, hypopnea index; RDI, respiratory disturbance index; RERA, respiratory effort related arousal; PLMS index, periodic leg movements in sleep index; Mean SaO2, mean oxygen saturation.

Table 2
PSG results of treatment with PMFT (with oral device).

N = 48 (completed 1 year research)	(1) PSG Pre Mean ± SD	(2) PSG-0.5 y Mean ± SD (with oral device)	(3) PSG-1y Mean ± SD (without oral device)	p-value	Post hoc
Body Hight (cm)	129.62 ± 22.23	137.04 ± 20.38	10.88 ± 20.13	<0.001***	1 < 2<3
BMI	17.04 ± 3.05	18.29 ± 3.72	18.53 ± 3.99	0.151	
AHI in sleep (event/hr)	6.00 ± 7.23	2.64 ± 2.47	2.44 ± 2.28	0.001*	1 > 2,3
AHI in REM (event/hr)	9.69 ± 11.22	7.26 ± 9.76	5.65 ± 6.92	0.023*	1 > 3
AI (event/hr)	1.51 ± 2.49	0.89 ± 1.02	0.79 ± 0.83	0.104	
HI (event/hr)	3.70 ± 4.70	1.75 ± 2.03	1.73 ± 2.01	0.001*	1 > 2,3
Desaturation Index	4.30 ± 7.49	2.65 ± 3.48	2.14 ± 1.87	0.125	
RDI (event/hr)	8.09 ± 13.85	6.32 ± 4.83	4.52 ± 2.99	0.330	
Efficiency %	84.42 ± 12.34	87.50 ± 10.32	89.99 ± 8.62	0.044*	
Awake %	11.26 ± 9.32	7.65 ± 7.36	11.25 ± 27.30	0.585	
REM %	20.52 ± 5.43	19.13 ± 5.80	18.61 ± 5.89	0.109	
Stage 1%	10.41 ± 7.90	8.99 ± 5.66	10.58 ± 5.72	0.297	
Stage 2%	42.16 ± 8.87	44.63 ± 8.39	44.46 ± 8.78	0.278	
Stage 3%	25.88 ± 9.38	27.22 ± 8.43	26.32 ± 8.48	0.675	
Total sleep time (mins)	382.51 ± 65.37	387.96 ± 66.89	391.43 ± 45.09	0.629	
Awake Count	19.75 ± 11.37	17.48 ± 11.97	17.76 ± 6.67	0.450	
Sleep Latency (mins)	13.68 ± 10.80	14.62 ± 17.70	11.23 ± 11.71	0.429	
REM Latency (mins)	103.81 ± 70.02	98.14 ± 58.32	94.06 ± 37.43	0.694	
Arousal Index (event/hr)	12.09 ± 9.45	9.61 ± 5.50	10.53 ± 6.23	0.094	
Mean Heart Rate	74.64 ± 13.54	69.20 ± 15.29	71.17 ± 9.00	0.070	
PLM Index (event/hr)	0.86 ± 2.87	0.61 ± 1.73	0.66 ± 2.16	0.868	
Snore Index (event/hr)	212.91 (271.95)	82.02 (126.98)	83.16 (144.13)	0.046*	1 > 2,3
Obstructive Apnea	3.08 ± 8.72	1.08 ± 3.68	0.95 ± 2.01	0.128	
Central Apnea	3.97 ± 6.17	3.59 ± 4.04	3.54 ± 4.08	0.833	
Mixed Apnea	1.44 ± 4.35	0.54 ± 1.29	0.18 ± 0.56	0.137	
Hyponea	18.36 ± 23.46	9.92 ± 11.53	9.74 ± 11.51	0.006**	1 > 2,3
RERA	19.37 ± 23.22	19.37 ± 20.32	19.40 ± 24.24	1.000	
Mean SaO2%	97.36 ± 0.67	96.75 ± 4.78	97.57 ± 0.68	0.365	
WASO	31.28 ± 27.24	29.05 ± 30.30	24.63 ± 21.62	0.486	

ANOVA, *P,0.05, **p < 0.01; Post hoc: Bonferroni test.
AHI, apnea-hypopnea index; AI, apnea index; HI, hypopnea index; RDI, respiratory disturbance index; RERA, respiratory effort related arousal; PLMS index, periodic leg movements in sleep index; Mean SaO2, mean oxygen saturation.
PSG-0.5 y: PSG was performed with usage of oral device at time of recording; PSG-1 y: PSG was performed without usage of oral device at time of recording.

Table 3
Comparison of the difference of PSG outcome between the treatment with MFT and PMFT (with oral device).

	MFT (0.5 y-pre) Mean ± SD (n = 23)	PMFT (0.5 y-pre) Mean ± SD (n = 52)	P1-value	MFT (0.5 y-pre) Mean ± SD (n = 10 with good compliance)	PMFT (0.5 y-pre) Mean ± SD (n = 48 with good compliance)	P2-value
BMI	0.13 ± 1.26	0.67 ± 2.21	0.185	0.64 ± 1.09	0.40 ± 2.13	0.605
AHI in sleep	−0.29 ± 1.38	−3.28 ± 4.51	<0.001*	−0.41 ± 0.18	−3.36 ± 4.05	**<0.001***
AHI in REM	−1.32 ± 5.52	−2.06 ± 6.57	0.595	−1.75 ± 3.04	−2.34 ± 4.02	0.601
AI	−0.38 ± 1.08	−0.71 ± 1.78	0.327	−0.51 ± 1.08	−0.62 ± 2.36	0.820
HI	0.08 ± 1.12	−2.27 ± 3.30	<0.001*	−0.29 ± 0.46	−2.95 ± 3.96	**0.001***
Desaturation Index	0.29 ± 2.32	−1.27 ± 3.12	0.019*	−0.65 ± 0.78	−1.56 ± 4.61	0.205
RDI	−0.55 ± 1.46	−1.93 ± 10.87	0.372	−0.59 ± 1.23	−2.29 ± 11.27	0.314
Efficiency %	0.31 ± 5.78	3.04 ± 1.82	0.030*	0.64 ± 4.63	3.08 ± 1.16	0.103
Awake %	1.05 ± 5.40	−4.40 ± 6.65	<0.001*	1.81 ± 3.40	−4.64 ± 6.42	**<0.001***
REM %	−2.74 ± 8.51	−1.80 ± 5.42	0.627	−5.24 ± 4.17	−2.39 ± 5.22	0.066
Stage 1%	3.59 ± 8.39	−0.43 ± 6.20	0.043*	1.66 ± 5.84	−1.42 ± 6.48	0.142
Stage 2%	0.65 ± 7.19	4.97 ± 8.91	0.030*	0.93 ± 5.79	2.47 ± 11.29	0.532
Stage 3%	−1.37 ± 7.08	0.76 ± 8.74	0.268	−3.74 ± 7.15	1.34 ± 9.83	0.062
Total sleep time (mins)	−1.28 ± 27.10	3.33 ± 15.23	0.447	−7.50 ± 21.61	5.45 ± 9.21	0.068
Sleep Latency (mins)	−5.81 ± 12.20	4.10 ± 15.34	0.004*	−4.07 ± 10.13	6.45 ± 15.49	**0.009***
Arousal Index	−0.16 ± 2.53	−2.67 ± 5.64	0.010*	−0.79 ± 2.04	−2.48 ± 4.83	0.081
Mean Heart Rate	−4.56 ± 10.74	−6.02 ± 14.10	0.459	−2.31 ± 13.15	−5.44 ± 16.68	0.517
PLM Index	0.81 ± 2.90	−0.40 ± 2.26	0.080	0.95 ± 2.56	−0.25 ± 2.86	0.192
Snore Index	−31.69 ± 108.65	−36.73 ± 177.51	0.881	−21.39 ± 161.61	−32.67 ± 183.04	0.845
Obstructive Apnea count	−0.06 ± 0.93	−2.00 ± 7.87	0.084	−0.16 ± 0.85	−2.00 ± 8.00	0.126
Central Apnea count	−1.75 ± 7.13	−0.29 ± 6.05	0.395	−3.77 ± 7.98	−0.38 ± 6.85	0.216
Mixed Apnea count	−0.44 ± 2.19	−0.87 ± 4.77	0.594	−0.31 ± 1.14	−0.90 ± 2.70	0.271
Hyponea count	0.25 ± 6.64	−8.41 ± 22.80	0.014*	0.08 ± 4.15	−8.44 ± 20.81	**0.012***
RERA	−1.63 ± 3.83	−0.95 ± 21.86	0.829	−1.05 ± 3.27	−0.97 ± 24.39	0.983
Mean SaO2%	−0.61 ± 4.92	−0.72 ± 4.33	0.992	−0.08 ± 0.43	−0.63 ± 3.83	0.338

P1 and P2: Intergroup comparison (T-Test for difference between groups).
AHI, apnea-hypopnea index(event/hour); AI, apnea index(event/hour); HI, hypopnea index(event/hour); RDI, respiratory disturbance index(event/hour); RERA, respiratory effort related arousal(event/hour); PLMS index, periodic leg movements in sleep index(event/hour); Mean SaO2, mean oxygen saturation.
Good compliance: compliance of treatment>80%.

Table 4
Comparison of the difference of cephalometric outcome between the treatment with PMFT (with Oral device) and MFT.

	PMFT (Oral device) (n = 48)			MFT (n = 10)			Before and after change		
	Ceph-pre Mean ± SD	Ceph-0.5 y Mean ± SD	p value	Ceph-pre Mean ± SD	Ceph-0.5 y Mean ± SD	p value	PMFT 0.5 y-pre Mean ± SD	MFT 0.5Y-Pre Mean ± SD	**P_T value**
Age	7.97 ± 3.08			7.10 ± 2.15					0.358
Gender (Male %)	64.6%			40.0%					0.043*
AHI event/hr	6.00 ± 7.23	2.64 ± 2.47	0.023*	2.46 ± 1.52	1.46 ± 1.41	0.015*	−3.36 ± 4.05	−1.00 ± 1.18	0.008*
ANB	4.14 ± 2.49	4.29 ± 1.84	0.645	6.67 ± 2.16	6.60 ± 2.02	0.779	0.16 ± 1.79	−0.07 ± 0.82	0.705
PNS-NPhp	16.90 ± 5.29	16.58 ± 4.90	0.680	11.64 ± 3.81	13.66 ± 3.81	0.028*	−0.32 ± 3.83	2.02 ± 2.45	0.063
PMm-NPh	9.48 ± 0.77	9.76 ± 0.58	0.663	6.40 ± 3.30	7.55 ± 3.00	0.103	0.28 ± 3.37	1.15 ± 1.82	0.444
OPha-OPhp	11.14 ± 2.30	12.17 ± 0.59	0.127	11.87 ± 5.67	9.63 ± 2.07	0.241	1.03 ± 3.55	−2.25 ± 5.51	0.036*
MinRGA	11.52 ± 0.48	14.02 ± 0.57	0.001***	12.87 ± 3.33	11.99 ± 2.20	0.508	2.50 ± 3.61	−0.88 ± 3.25	0.013*
PNS-AD2	13.43 ± 0.74	14.71 ± 0.75	0.027*	11.15 ± 2.58	13.23 ± 2.99	0.028*	1.28 ± 2.95	2.09 ± 2.23	0.434

ANB:A point-nasion-B point angle (normal range is about 2–4); PNS− NPhp:Distance between PNS and posterior side of nasopharynx; PMm-NPh: Distance soft palate-posterior side of nasopharynx; OPha-OPhp: Distance anterior side-posterior side of oropharynx. MinRGA: Minimal retroglossal airway. PNS-AD2: Distance from PNS to the nearest adenoid tissue measured along the line perpendicular to S-BA.
Oral device significantly improved the width of airway at the level of nasopharynx (MinRGA, PNS-AD1, PNS-AD2).
P_T: Intergroup comparison: Mann−Whitney test for difference between groups.
P: Intragroup comparison: Wilcoxon signed-rank test.

good compliance (n = 10) showed also significant improvement in PNS-NPhp and PNS-AD2 measurements. It is obvious that the improvement of AHI and upper airway of group B (passive MFT) is more than group A (MFT) (Table 4).

Moreover, at the one year follow-up of cephalometric data in group B (passive MFT) showed ANB: 3.8 ± 2.5 mm (p = 0.482); Distance between PNS and posterior side of nasopharynx (PNS-NPhp): 18.1 ± 4.2 mm (p = 0.144); Distance soft palate-posterior side of nasopharynx (PMm-NPh): 10.52 ± 2.1 mm (p = 0.095); Distance anterior side-posterior side of oropharynx (OPha-OPhp): 11.8 + 3.6 mm (p = 0.155); MinRGA: 13.5 ± 3.6 mm (p = 0.010); PNS-AD1: 17.0 ± 3.7 mm (p = 0.001); and PNS-AD2: 17.0 ± 3.7 mm (p = 0.001). The one year treatment with oral device significantly improved the width of the airway also.

5. Discussion

Prior publications looking at role of MFT in improving sleep related breathing had emphasized the very large amount of non-usable data [10–15]. Compliance with treatment is a major issue. In prior studies we learned that it is very important, particularly at beginning of treatment to see child and parents several times a week and to involve in depth one of the parents that will perform the exercise in same time as child. Such involvement may be costly depending of the social-security system and the reimbursement provided for the specialist involvement. But this is not the only issue: training was free in our study. The daily involvement of at least one parent with performance of the exercises at the same time in the morning and/on evening with the child is a critical issue. Social

factors are important in MFT: often both parents work outside; families may have several children; work and school schedules, and-particularly in Far East Asian countries-with the very large demands from schools and very demanding home-work time are clear handicap to parental involvement in daily reeducation programs. Even in the 10 families where good compliance was noted at 6 months, we were unable to pursue our long term follow-up. Once parents learned that there were positive results at 6 months, they stopped MFT, and we were unable to appreciate if there was persistence of positive effect at 12 months. Functional appliances have been tried for many years the latest reports by orthodontists and recently by sleep-specialists as example Vila et al. [17] and Chen et al. [18], but the presented device is different in as much as it aims at lifting the tongue upward aiming to have simultaneous contraction of tongue muscles. The "passive MFT" requires very little involvement of parents, and children are very quickly used of having the device in their mouth, so compliance is not a problem. But placing any dental device in a mouth of a child is always an issue: Any effect induced on one of the jaw will lead quickly to a compensatory effect on the other-one. Many devices are advertise for children, but very little, if any, study has been performed to appreciate the long term-changes induces on upper and lower jaws when an appliance is used for several months, despite the fact that such changes are always noted to some degree after months of usage. We kept our device for one year, and could not see any change at our clinical and imaging evaluations. But we used cephalometrics-X-rays and probably in the future 3D-dental CT will be a better tool. Usage of such devices as ours must be done with clear follow-up and usage must be with clear time limit. Also long term follow-up without any of these devices is an important issue.

How the device does really works, has not been completely worked-out. It is a type of "functional appliance", and functional appliances have been "advertised" for over 70 years [17,18], as having effects on oro-facial growth, but the secondary effects noted when usage is too long and the unavoidable impact on the other jaw has been clear limitation in their usage.

One crucial finding with the PSG at 1 year post-recording compared to baseline is that, without the device children, breathed through their nose during sleep, while mouth breathing was the rule at baseline including in our post T&A children. All night nasal breathing is the only demonstration of successful treatment of the upper-airway [19–22].

To sum up, our study is the first prospective study performed on a randomized group of children with pediatric OSA that demonstrate that MFT can lead to nasal breathing during sleep. It also outlined the very important difficulties associated with such treatment. The limitations are the high drop-out rate of MFT group and we only followed one year side effect of oral device treatment (PMFT). This study indicates that "passive MFT" may be a valid alternative, but very careful attention to the potential negative secondary impact of the device, that may not be immediately appreciated, is a clear requirement.

Author contributions

Study conception and design: Y-S Huang, T Paiva and C Guilleminault. Acquisition of data: L-C Chuang, and Y-S Huang. Analysis and interpretation of cephalometric data: Michele Hervy-Auboiron and C-H Lin. Drafting of manuscript: Y-S Huang and C Guilleminault. Critical revision: Y-S Huang and C Guilleminault.

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Conflict of interest

None of the authors has conflict of interest to disclose.

All authors approved the final manuscript and agree to be accountable for all aspects of the work.

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Conflicts of interest

The author declares no conflict of interest.

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