Efetividade em Longo-Prazo de Dois Aparelhos Intraorais no Tratamento da Apneia Obstrutiva do Sono: Relato de um Caso

Long-Term Effectiveness of Two Oral Appliances for Obstructive Sleep Apnea: a Case Report

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Resumo

A síndrome da apneia obstrutiva do sono - SAOS é um distúrbio respiratório caracterizado por episódios recorrentes de obstrução das vias aéreas superiores durante o sono. Aparelhos intraorais - AIOs têm sido utilizados em pacientes com SAOS moderada ou severa que não se adaptaram ou recusam o tratamento com pressão positiva contínua nas vias aéreas - CPAP ou pacientes com impossibilidade de realização de cirurgia. Aparelhos de avanço mandibular têm sido amplamente utilizados com eficácia. Além de estabilizar a mandíbula, alguns AIOs permitem que o paciente faça movimentos mandibulares de lateralidade e verticais sem desencaixar o aparelho, reduzindo o risco de lesionar a articulação temporomandibular. O objetivo desse estudo foi avaliar a efetividade de dois tipos de AIO no tratamento da apneia. Este estudo apresenta o caso de um paciente com SAOS moderada e grave dessaturação de oxigênio (SaO₂ mínima de 55%), tratado com dois tipos de aparelho intraoral: o PM PositionerTM, que não permite movimentos laterais da mandíbula, e a Placa Lateroprotrusiva (PLP®), que permite movimentos de lateralidade. O aparelho PLP® foi mais efetivo se comparado ao PM Positioner. Através da avaliação em longo prazo observouse que o PLP® proporcionou mais conforto ao paciente, maior aderência ao tratamento e uma maior capacidade de avanço mandibular, quando comparado ao aparelho que não permitia movimentos mandibulares de lateralidade.

Palavras-chave: Apneia do Sono Tipo Obstrutiva. Avanço Mandibular. Desempenho de Aparelho Ortodôntico.

Abstract

Obstructive sleep apnea syndrome (OSAS) is a breathing disorder characterized by recurrent episodes of upper airway obstruction during sleep. Oral appliances have been used in patients with moderate to severe OSAS, who cannot tolerate or refuse the therapy with continuous positive airway pressure or candidates who present impossibility of performing surgery. Oral appliances such as mandibular advancement devices (MADs) have been widely used and proven to be effective. In addition to stabilizing the mandible, some MADs allow the patient to move it laterally and vertically without disengaging the appliance, reducing the risk of injuring the temporomandibular joint. The aim of this study was to evaluate the effectiveness of two types of oral appliances in the treatment of apnea. A patient who presented moderate OSAS and severe oxygen desaturation (SaO $_2$ minimum of 55%) was treated by two different types of MADs: the PM Positioner TM , which is a device that do not allow lateral movements of the mandible, and the Placa Lateroprotrusiva (PLP $^{\otimes}$), which allows lateral movements. The PLP $^{\otimes}$ was more effective than the PM Positioner TM . Long-term assessment revealed that PLP $^{\otimes}$ was more effective because it provided more comfort and a greater capacity for mandibular advancement, when compared to a device which did not allow the jaw to move laterally.

Keywords: Sleep Apnea. Obstructive. Mandibular Advancement. Orthodontic Appliance Design.

1 Introduction

Obstructive sleep apnea syndrome - OSAS is a breathing disorder characterized by recurrent episodes of partial or complete obstruction of the upper airway during sleep, which leads to intermittent hypoxemia, transient hypercapnia and frequent arousals, associated with signs and/or clinical symptoms¹. The signs and symptoms of OSAS are commonly described as excessive sleepiness, snoring, presence of respiratory pauses during sleep, cognitive impairment, cardiovascular disease, anxiety, depression and metabolic dysfunction²⁻⁸.

Polysomnography is usually required when diagnosing OSA among adults, once it indicates the disorder's severity, and supports the treatment planning¹. The classification of

OSAS's severity depends on both the degree of daytime sleepiness and the apnea-hypopnea index - AHI, which refers to the total number of complete cessations (apnea) and partial obstructions (hypopnea) of breathing occurring per hour of sleep^{1,5,9,10}.

A recent study in Brazil has shown that 55% of the population of São Paulo suffers from sleepiness, fatigue (38.9%), snoring (20.5%), while 29.2% reported breathing interruption. The research found an average of 32.8% of the participants with OSAS⁵.

Oral appliances are one of the clinical approaches for the treatment of OSAS¹¹. They are used during sleep to prevent both the tissues of the oropharynx, and the base of the tongue from collapsing and causing airway obstruction¹²⁻¹⁴. The

OAs are indicated for patients who present primary snoring or have mild OSAS, who do not respond or are not suitable for treatment with behavioral measures such as weight loss or changes in sleep position. Moreover, OAs are advised for patients with moderate to severe OSAS who cannot tolerate or refuse the continuous positive airway pressure - CPAP therapy, or candidates who present impossibility of performing surgery^{11,15,16}.

Basically, there are four models of OAs: tongue retainers devices (TRD), mandibular advancement devices - MADs, adjustable soft palate lifter – ASPL, and oral positive airway pressure devices¹⁷⁻¹⁹. However, most devices are designed to be MADs²⁰, and presented the best results when treating OSAS^{20,21}, once they modify the upper airway by changing the posture of the jaw and tongue^{8,11,22,23}.

The category of MADs can be subdivided into adjustable or non-adjustable and classified according to the manufacturing method; prefabricated or made in the laboratory. The material used in manufacturing can be hard or soft, and the retention can be in the maxilla or in both arches^{15,24}. Besides stabilizing the jaw, some MADs allow lateral movements, retraction, protrusion and opening up, therefore they reduce the risk of injuring the temporomandibular joint - TMJ, increasing comfort and the patient adherence to treatment²⁵⁻²⁷.

There is a relatively limited number of trials comparing the efficacy of customized OAs designs. Nevertheless, the literature suggests that different designs are similarly effective in treating OSA²⁸. This study presents a case of a patient with moderate OSAS (16.2 / h), with significant oxygen desaturation (SaO₂ minimum of 55%) treated by two types of MADs: the PM PositionerTM ²⁹ and *Placa Lateroprotrusiva* (PLP®). The patient was assessed and monitored in the long term. An informed consent form explaining the purpose of the study and requiring the written consent was given to the patient. The patient agreed with the study and the consent form was properly signed.

2 Case Report

The patient, female, 59 years old, was referred by an otorhinolaryngologist after the resection of enlarged uvula. The patient's main concern was to reduce the persistent fatigue and sleepiness. During anamnesis, the patient reported having already used an oral appliance for three years, but had to discontinue the treatment due to joint pain. The patient presented Epworth sleepiness score³⁰ of 14, and body mass index (BMI)³¹ of 31.2 Kg/m².

2.1 Clinical examination

During dental examination, the morphological analysis of the face in frontal and lateral views was classified as pattern I, which is characterized by the normal sagittal and vertical skeletal relationships. The patient had apparent symmetry in front view, and presented convexity in the face similar to the pattern I of normality in lateral view³². The mandibular movement was performed in a coordinated manner and the intraoral examination showed a mandibular midline shift to the left, of 1 mm, Class III canine relationship on the left side, and Class I on the right³³. According to Angle's classification³³, the patient presented an occlusion, classified as class I or neutrocclusion on both sides. A posteriorized soft palate¹⁰, macroglossia¹⁰, Mallampati grade IV¹⁰, and tonsil grade II was also detected ¹⁰.

3 Results and Discussion

3.1 Polysomnography

AHI index was defined as the number of episodes of apnea plus episodes of hypopnea per hour of sleep¹⁰. OSA was defined as AHI > 5.40. The baseline polysomnography showed an apnea index (AI) of 9.7 / h, hypopnea index (HI) of 6.5 / h, with an AHI of $16.2 / h^{10}$. The average oxygen saturation was 86% and minimum 55%¹⁰. The percent of total sleep time below 90% saturation was 99.5%, a sleep efficiency of 81%, N3 sleep time of 7%, REM of 18% and the awakening index of $35.6 / h^{10}$.

3.2 Radiographic analysis

Digital panoramic radiographic analysis revealed absence of carious lesions. There were 28 erupted teeth and one impacted tooth (lower right 3rd molar), absence of upper 3rd molars and lower left 3rd molar. It was also noticed the absence of apical lesions, thickening or enlargement of the periodontal space. There was generalized loss of interdental bone crest (maxilla and mandible). Condyles and condylar eminences were normal³⁴.

3.3 Cephalometric analysis

Since the 1980s, cephalometry can be used as a complementary test in patients with OSAS to assist in the identification of craniofacial anatomical determinants involved in pharyngeal collapse during sleep. The cephalometry in lateral view is relatively easy to analyze, low-cost, and emits minimal radiation levels, offering a two-dimensional visualization of anatomic structures³⁵.

Despite the difficulty of comparison, some characteristics are typical in patients with OSAS. These characteristics include the increase in the length of the soft palate, thicker soft palate, reduced amplitude of the pharyngeal airway space, micrognathia, retroposition of the maxilla or mandible, and alterations in the hyoid bone position. Some other anatomical factors may contribute to the obstructive process of the upper airway during sleep, including macroglossia, tonsillar hypertrophy and tumors in the upper airway³⁶.

The patient's cephalometric evaluation (Figure 1) provided information of various anatomical regions that maintain close correlation with obstructive sleep-disordered breathing. The results are listed in Table 1.

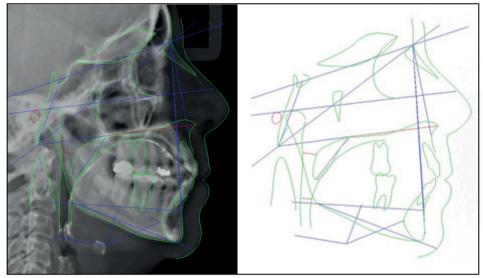


Figure 1: Patient's cephalometric lines, planes and angles evaluated in the present study.

Table 1: Summary of cephalometric measurements

Cephalometric variables	Baseline values	Normal
SNA (°)	82.7	82.00 <u>+</u> 2.00
SNB (°)	75.2	80.00 ± 2.00
ANB (°)	7.5	2.00 <u>+</u> 1.00
SN-MP (°)	39.6	32.00 <u>+</u> 3.00
FMA (°)	27.8	25.00 <u>+</u> 4.00
Ba-SN (°)	128.6	130.00 <u>+</u> 5.00
PFH (mm)	85.7	88.00 <u>+</u> 4.00
AFH (mm)	136.9	136.00 <u>+</u> 6.00
PFH – AFH (%)	62.6	0.00
Ba- PNS (mm)	48.6	48.00 <u>+</u> 4.00
SPAS (mm)	9.9	11.00 <u>+</u> 3.00
PAS (mm)	6.9	11.00 <u>+</u> 2.00
PNS-P (mm)	37.1	37.00 <u>+</u> 3.00
SPL (mm)	11.7	11.00 <u>+</u> 2.00
MPH (mm)	19.2	15.00 ±3.00
H-C3 (mm)	35.9	40.00 <u>+</u> 5.00
H-RGN (mm)	42.6	41.00 <u>+</u> 8.00

PFH = posterior facial height; AFH= anterior facial height; PNS= posterior nasal spine; SPAS= superior posterior airway space; PAS= posterior airway space; PNS- P= posterior nasal spine to tip of palate; SPL= soft palate length; MPH= mandibular plane-hyoid distance; H-C3= Distance between the third cervical vertebrae and the hyoid bone; H-RGN= distance between the hyoid bone and retrognathic

When comparing to normal measurements³⁶, the patient presented an increased ANB value (7.5°), indicating a possible mandibular retroposition. The SN-MP was also increased, and a correlation between posterior facial height - PFH and anterior facial height - AFH below 65% was observed, suggesting a clockwise facial growth. The posterior airway space - PAS decreased when compared to the normal pattern³⁶. The soft palate length - SPL and the maximum palate thickness - TPS

exhibited normal patterns³⁶. It was also identified that the distance from the hyoid bone to the mandibular plane - MPH was slightly increased, indicating a small displacement of the hyoid bone, despite the normal findings ³⁶ in anteroposterior position (H-C3 and H-RGN) (Table 1).

It was found 81.66 mm for a standard of 72.50 ± 3.00 of Tongue-Length and 25.78 mm for a standard 24.00 + 3.00 of Tongue-Height, indicating that the patient has a greater lingual length when compared to the standard measurements³⁷.

3.4 Oral appliances

Concerning the mechanism of action, both oral appliances were mandibular advancement devices used to modify the position of upper airway structures in order to enlarge the airway⁸.

- *PM Positioner* ^{™ 29}: The device has plates that fit over all upper and lower teeth. Expansion screws are located in the left and right mouth areas to allow maximum space for the tongue and favor the forward positioning of the jaw for maximum effectiveness. The clamps are placed internally to the plates for better retention of the device. Forty-four forward positions are available in increments of 0.25 mm, covering 11.0 mm range of anteroposterior movement²⁹ (Figure 2).



Figure 2: Mandibular advancement device: *PM Positioner* TM.

- Placa Lateroprotrusiva- PLP ® Device: This device was developed by one of the authors (Dr. Guimarães), and consists of two encapsulated acrylic plates retained by the two arcades through retaining clamps. The plates are lateroposteriorly joined together by accessories that enable lateral movements and progressive advancement of the mandible. These accessories are made of screw expanders that will receive barcode orientation drive.

The screws are available in increments of 0.25 mm, comprising a total of 11 mm in anteroposterior movement. The guide bars have a bayonet shape and their tips are held to the acrylic plates with the aid of telescopic tubes. These tubes allow the movement of laterality. The configuration of the bars follows this format so they can pull the plates in an effective manner, allowing the progressive advancement of the mandible.

The plates have acrylic tracks that allow progressive advancement and laterality movement of the jaw, which are realized in a stable, solid, and interference-free path. Although such tracks have the purpose of guiding the movement, they have no function in relation to existing orthopedic devices (Figure 3).



Figure 3: Mandibular advancement device: *Placa Lateroprotrusiva*-PLP®.

The patient's treatment was based on the recommendations proposed by the American Academy of Sleep Medicine³⁸. To obtain the maximum protrusion and bite registration for the oral appliance manufacture, the George Gauge device ® was used. The maximum protrusion was of 12 mm, and the therapeutic protrusion was calculated at 70% (8.4 mm) of this measure. The PM Positioner TM ²⁹ device was the first choice for the patient's treatment. The OA was made in maximum habitual intercuspation - MHI due to the patient's history of joint pain while making use of OA.

The vertical height was 2mm, considering the patient facial type. After installing the device, the patient underwent four visits for protrusion adjustments, reaching the maximum of 6 mm of adjustment due to complaints of mild pain in the TMJ. The patient was then referred for overnight

polysomnography with the OA for therapeutic control. Despite the results showed no improvement in AHI, an improvement in oxygen saturation was observed (93.1%).

The PM PositionerTM device was used for 8 months. Nevertheless, the control polysomnography did not present satisfactory results, therefore after a two-weeks interval of wash-out^{39,40} this device was substituted by an OA that enabled laterality – the PLP [®]. After installing this device, the patient underwent for four clinical visits, and the protrusion achieved was of 8 mm, and again, the patient was referred for an overnight polysomnography with the oral appliance. The patient underwent regular dental follow-up visits every six months as well as control polysomnographys, on November, 2007 and August, 2011 (Table 2).

Table 2: Polysomnography results in relation to time and type of oral appliance used.

Variables	Baseline	Oral appliance PMP TM	Oral appliance PLP®	Oral appliance PLP®
Date (month/day/year)	09/06/2003	06/20/2004	11/11/2007	08/31/2011
AHI (/h)	16.2	16.4	8.9	5.6
AI (/h)	9.7	1	1.6	0.1
HI (/h)	6.5	15.4	7.3	5.4
Min Sa O ₂ (%)	55	72	83.9	88
Sa O ₂ (%), TST < 90%	99.5	7.3	48	1
Sleep efficiency (%)	81	95.9	78	89.3
Stage N3 (%)	7	0	5	4.7
REM sleep (%)	18	19.7	22	23.9
Arousal index (/h)	35.6	3	12	8.7
BMI (Kg/m²)	31.2	31.2	27.5	27.0

AHI= apnea-hipopnea index; AI = apnea index; HI= hypopnea index; SaO_2 = oxygen saturation by pulse oximetry; TST= Total sleep time (minutes); REM= rapid eyes movement; BMI=body mass index.

By the end of the study, the patient continued reporting improvements in the sleep quality, and was feeling more prepared for daily activities, with no daytime sleepiness. No pain in the masticatory muscles and TMJ was reported.

Both OAs were made with hard acrylic. Toothaches caused by devices made with hard acrylic are indicative of a defect in its construction, which can be eliminated by adjusting the plate in the corresponding area of the affected teeth²⁶.

The need for successful management of treatments with OA in the long term has been stated in literature⁴¹ The efficacy of the patient's treatment from September 2003 to August 2011 revealed that the patient's respiratory parameters were stable and controlled, as well as the subjective reports of restful

sleep and no drowsiness. The patient of the present study had moderate OSAS, and clinical characteristics favorable to the use of OA. The results are consistent, with evidence of improvement in subjective sleepiness and respiratory disturbance indexes presented by a Cochrane review⁴¹.

A study published in 2000 reported no adverse effects after two years of use of a monoblock³⁷. However, some authors have mentioned that rigid OAs that do not allow movements of laterality may cause more TMJ dysfunction, leading to less compliance by patients²⁶, which is consistent with the findings in the present case report.

During the treatment with the PM Positioner™, the patient complained of pain in TMJ and muscles, which limited the titration of the OA. Data from the first polysomnography showed significant improvement in oxygen saturation and apnea index, but not in the AHI. Therefore, after using the first OA for 8 months, the device was replaced by the PLP®, which allows lateral mandibular movements and provides greater comfort and less facial pain, enabling greater mandibular advancement.

Nocturnal oxygenation normalization depends significantly on the body weight and the severity of desaturation before treatment⁴². Moreover, the mandibular advancement results in dose-dependent reduction of closing pressure of the pharynx^{43,44}. Successful nocturnal oxygenation improvement seems to be achieved when the OA reduces the pharynx closing pressure below atmospheric pressure, and each 2 mm of mandibular advancement coincides with approximately 20% improvement in the quantity and severity of nocturnal desaturation⁴². The present data are in compliance with the previous studies, once greater normalization of oxygenation was found with major advances in the patient protrusion.

Although some studies have reported that mouth vertical opening should be the lowest possible, there is still no polysomnographic study establishing the optimal vertical opening between the incisors, but the most adopted opening measure is of 2 mm²⁶.

It is worth mentioning that possible collateral effects related to the use of oral appliances were not investigated. Therefore, the lack of cephalometric analysis and further evaluations for controlling the patient before and after the treatment with the OAs are limitations of the present study.

A recent review on the effectiveness of different devices found that when monoblock devices are compared with devices made by two pieces, no significant differences were observed regarding the reductions in AHI²⁰. Despite the PM Positioner TM is a two-piece appliance, it does not allow lateral movements, and in this case this characteristic resulted in limiting the degree of mandibular advancement due to joint/ muscle pain, while devices such as PLP ® allowing lateral movements provide greater comfort and better results concerning the reduction in AHI as well as improvements in oxygenation.

4 Conclusion

The choice related to the type of OA for OSAS treatment can influence the treatment effectiveness. In the present study, the OA that enabled lateral movements was more effective, once less discomfort and greater possibility of mandibular advancement was observed.

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