ISSNe 2178-1990

10.7308/aodontol/2018.54.e03

ARQUIVOS EM ODONTOLOGIA

Assessment of a novel type of oral appliance for the treatment of severe obstructive sleep apnea syndrome: a case series

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Aim: This study aimed to evaluate the respiratory parameters among patients diagnosed with severe obstructive sleep apnea (OSA) syndrome treated with a novel design of an oral appliance (OA): a lingual orthosis.
Methods: A case series study was conducted with 11 patients suffering from severe OSA, apnea-hypopnea index (AHI) > 30 events/h. All patients underwent pre- and post-treatment overnight polysomnography and were diagnosed by a physician based on polysomnography indexes. All selected patients had a history of nonadherence to continuous positive airway pressure devices.
Results: Before treatment, the mean AHI of the sample was 65.9 ± 30.5. After reaching the optimal OA titration, the polysomnography (PSG) showed an average AHI of 10.1 ± 5.5 ($p<0.001$). There was a significant reduction in obstructive events that decreased from 60.0 ± 31.7 to 8.5 ± 3.8 events/hour ($p<0.001$). The minimal oxyhemoglobin saturation showed significant improvement after the OA treatment, increasing from 74.3 ± 7.8 to 83.4 ± 4.9 ($p<0.05$).
Conclusion: Lingual orthosis was effective in resolving severe OSAS, showing satisfactory results within the present sample. Further studies are needed in order to broaden the knowledge regarding the effectiveness of this oral appliance.
Uniterms: Sleep apnea, obstructive. Orthotic devices. Case reports.

Submetido: 18/01/18
Aceito: 10/05/18

INTRODUCTION

The amount of systemic changes caused by obstructive sleep apnea (OSA), such as arterial hypertension, myocardial infarction and endocrine and psychological changes, has already been discussed in the literature[1,2]. The treatment of OSA aims to normalize sleep patterns and decrease awakenings associated with obstructive respiratory events. The gold standard treatment for OSA is the continuous positive airway pressure (CPAP) device[1,3]. Nonetheless, many patients do not adhere to or discontinue long-term treatment.[4,5] There are also surgical interventions; however, it has been shown that invasive treatments do not present consistent and lasting results in the treatment of severe cases[6].

Another treatment option is the oral appliance (OA), which is indicated for the treatment of mild and moderate OSA and can also be considered as a treatment option for patients suffering from severe OSA, when patients refuse CPAP therapy[3]. The majority

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of OA involves a mechanism of mandibular advancement, and there is abundant scientific evidence indicating their effectiveness in the treatment of OSA[4,7-14].

A novel design of OA proposed by Caram and Quintela (2013)[9], has been developed and studied in order to broaden patient's alternatives regarding OSA's treatment. Lingual orthosis has a mechanical direct action on the tongue, providing a lingual control, in turn avoiding the collapse of the tongue towards the posterior oropharyngeal structures.

The first report related to lingual orthosis showed significant results and improvements in respiratory parameters, such as the reduction in the apnea-hypopnea index (AHI). Since 1982, other oral devices, such as the Tongue Retainer, have already been implementing this mechanical action over the tongue[10]. The present study aimed to determine the effectiveness of a direct lingual control, provided by lingual orthosis, in the treatment of severe OSA.

MATERIAL AND METHODS

SAMPLE

The target sample was composed by 17 patients with severe OSA who do not adhere to CPAP treatment. Therefore, the patients were referred for treatment with the direct lingual control technique, using lingual orthosis (Figure 1).

To standardize the data collection, six patients were excluded from the main sample once their pre- and post-treatment polysomnography had been conducted in different sleep laboratories and/or full-night polysomnography (PSG) had not been conducted. This study was approved by the Research Ethics Committee of the University of São Paulo (USP), Brazil.

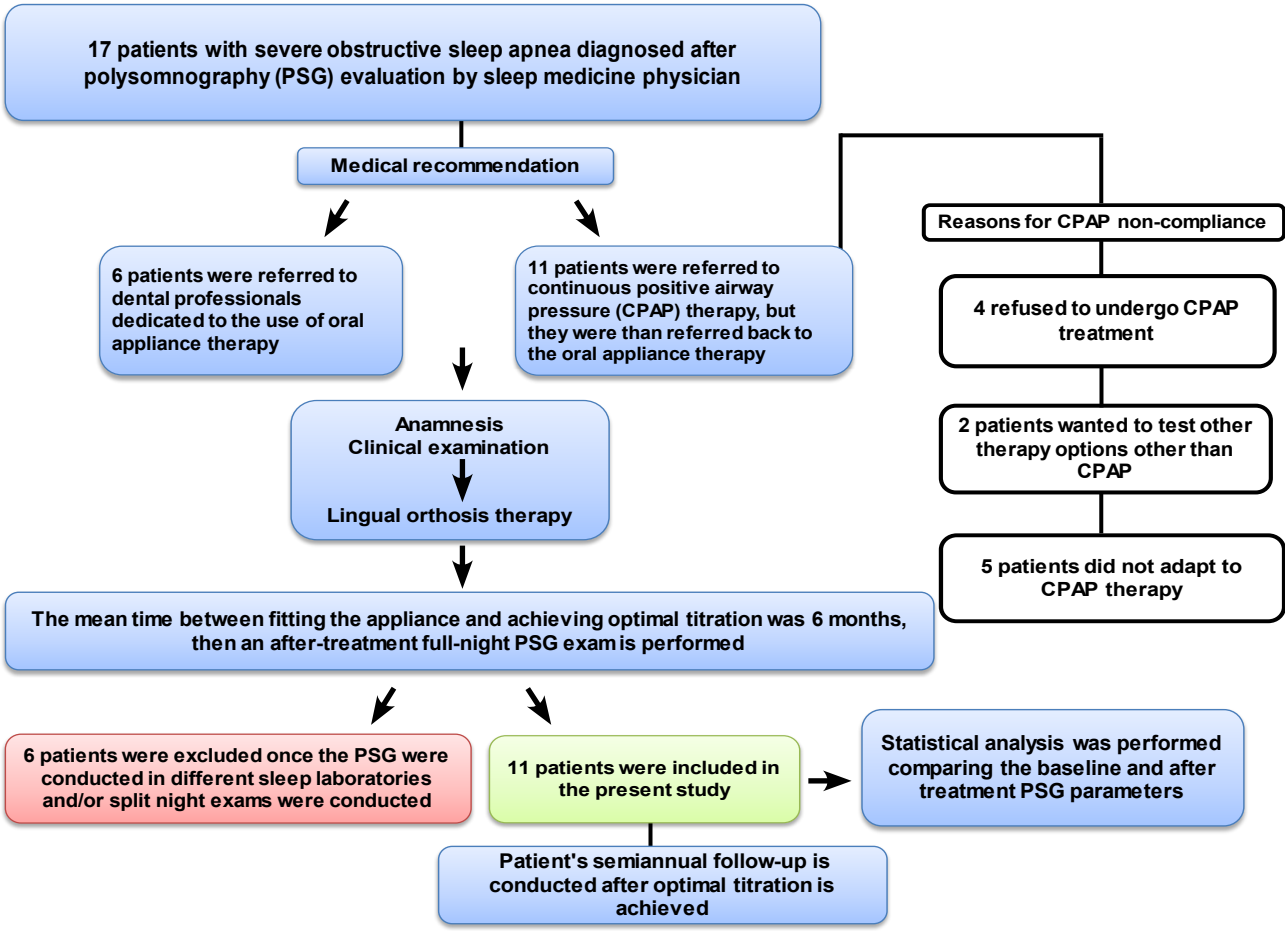

Figure 1 - Sample selection criteria and reasons for exclusion.

ANAMNESIS

During the clinical examination performed prior to treatment, some possible predictors of success were analyzed in each patient. Unlike mandibular advancement devices, the lingual orthosis does not require a considerable number of natural teeth for its stability, as it can be fitted in patients with partial or total prosthesis. In the present sample, the patients exhibited scores varying from 3 to 4, as defined by the Mallampati index, showing an increase in tongue volume,

which favors recommendations for the use of lingual orthosis. The index is an anesthetic assessment of the risk of intubation based on the anatomy of the oropharynx, and it has been suggested to be a possible simple assessment tool for OSA[3,6]. The assessment is divided into 4 classes, and each class is graded based on the visibility of the airway structures: Class I – pillars, tonsils, and soft palate are all clearly visible; Class II – the uvula, tonsil pillars, and upper pole are visible; Class III – only part of the soft palate and the base of the uvula is visible, and Class IV – only the hard palate is visible[15]. Daytime sleepiness and snoring were evaluated in all patients during the clinical examination.

POLYSOMNOGRAPHY EVALUATION

All patients were subjected to two full night polysomnography recordings: the first PSG was performed to diagnose sleep-related breathing disorders: the baseline recording. Once the OA was optimally titrated, resulting in a resolution of the symptoms, or the titration had achieved individual tolerance, the patient was referred to the sleep laboratory for polysomnography monitoring with the OA *in situ*. These exams allowed for the therapeutic effect of the titrated OA to be objectively assessed by comparing the PSG results.

The exams were conducted in sleep laboratories, monitored by trained professionals, and recorded at least for 6 hours. The parameters evaluated in this study included: electroencephalogram, electromyogram of the chin region, electromyogram of the lower limbs, nasal and oral airflow monitored by thermistor or thermocouple sensors, nasal pressure recording obtained by pressure transducer, recording of thoracic and abdominal movement through inductance straps and piezoelectric sensors, electrocardiogram, and digital oximetry. Snoring was evaluated with a tracheal microphone, and the body position was also recorded.

Sleep stages were assessed by the amplitude and frequency of the waves transmitted by electrodes. All sleep parameters were evaluated according to the standards established by the American Association of Sleep Medicine (AASM)[16]. The softwares used for data evaluation were “Alice Respironics” and “Somnologica”.

LINGUAL ORTHOSIS

This oral device was developed by Jorge Machado Caram in 1992 and was patented in 2009 (Patent number: PI 0101407-2). The lingual orthosis directly controls the tongue’s position, avoiding its collapse with the adjacent oropharyngeal structures. It consists of a jaw fitting single plate to which an acrylic lingual control device made of highly polished acrylic resin is attached (Figure 2). The lingual control appliance is adjusted according to the dental sleep criteria, aimed at providing the best risk-benefit ratio.

The titration of the lingual orthosis is done weekly, respecting the patient adherence and adaptation to the appliance. Sometimes biweekly or monthly titrations are necessary. At first, a small spring is placed and juxtaposed to the palate, which undergoes a process of stretching, in which it is directed towards the base of the tongue during the titration process. The device consists of two wires (caliber 1.0 mm) which are stretched in a ratio of 1 to 2 mm during each dental appointment. Nearly two months after beginning oral appliance therapy, the wires are fully extended and support the acrylic retainer that must be in close contact with the tongue’s base. Once some classic symptoms of OSA, such as excessive daytime sleepiness and snoring, have been treated, the patients are referred to a post-treatment PSG exam[17].

Figure 2 - The lingual orthosis.

IMAGING

To analyze the lingual orthosis' titrated position and the airspace obtained using the appliance, cephalometric radiography and computed tomography were requested for the entire sample (Figures 3 and 4).

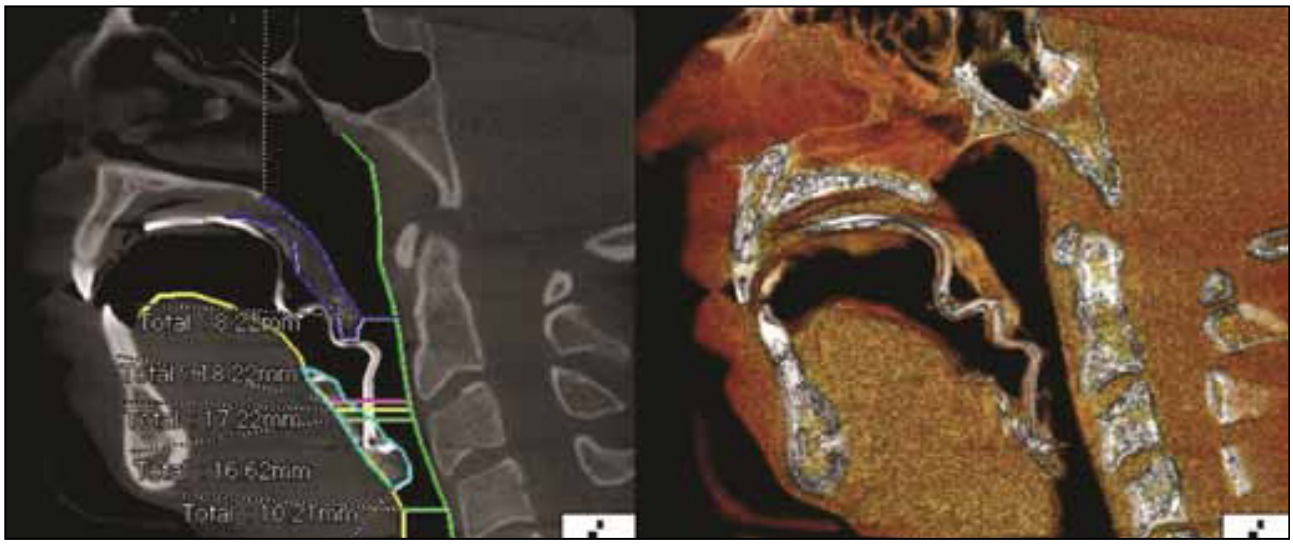

Figure 3 - Computed tomography. The sagittal plane shows Caram's lingual orthosis in situ. It can be observed a space between the soft palate and tongue position promoted by the oral appliance.

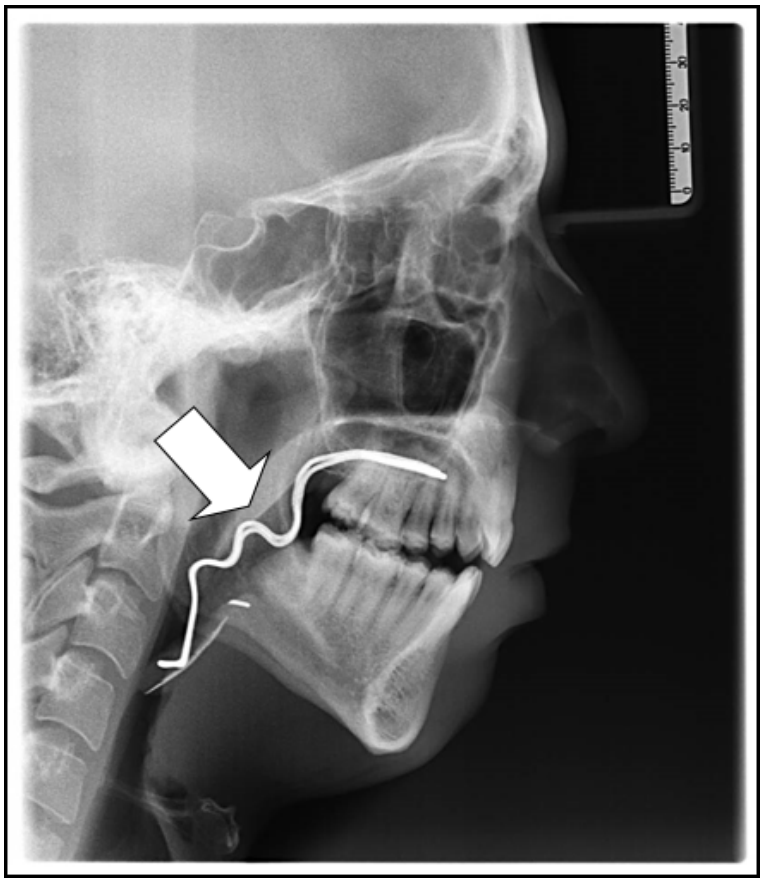

Figure 4 - Cephalometric radiography. The sagittal plane shows Caram's lingual orthosis *in situ*. It can be observed a space between the soft palate and tongue position promoted by the oral appliance (white arrow) and an increase in the oropharynx caliber.

STATISTICAL ANALYSIS

Statistical tests used univariate analysis by means of the Minitab© program. The level of statistical significance was 5%. Comparisons between baseline and after-treatment respiratory parameters were evaluated by the paired t-test. Following normal distribution, the data were measured by Mean and Standard Deviation.

RESULTS

Eleven patients took part in this study; the reasons for exclusion are shown in Figure 1. The sample's mean age was 55 ± 13 years, with 2 women and 9 men. Before treatment, the mean AHI of the sample was 65.9 ± 30.5. After reaching the optimal OA titration, the PSG showed an average AHI of 10.1 ± 5.5 ($p < 0.001$). A significant reduction in the obstructive events was observed, which decreased from 60.0 ± 31.7 to 8.5 ± 3.8 events/hour ($p < 0.001$).

Reduction in central events was also significant when compared to baseline events (decreasing from 2.0 ± 3.0 to 0.1 ± 0.3 events/hour, $p < 0.001$) and to mixed events (decreasing from 3.0 ± 3.2 to 0.1 ± 0.3 events/hour, $p < 0.001$). The minimal oxyhemoglobin saturation showed significant improvement after OA treatment, increasing from 74.3 ± 7.8 to 83.4 ± 4.9 (Table 1). Despite the benefits provided by the oral appliance, some transitory side effects were reported during the treatment, such as difficulty swallowing, gagging, and vomiting. These side effects occurred shortly after appliance titration. However, as the patient became more accustomed to the orthosis, these effects disappeared.

DISCUSSION

This study sought to present the medical and dental care community with more cases related to lingual orthosis, a novel therapeutic option among the arsenal of oral devices that deal with sleep-disordered breathing. In the present study, all patients underwent the baseline and after-treatment polysomnography in the same sleep laboratory in order to standardize the comparison of results. A recent study, published in 2018[17], also evaluated the efficacy of this appliance, but it focused on evaluating the impacts on central and mixed events. In addition, the polysomnographic exams were not conducted in the same laboratory.

Concerning the treatment of patients suffering from OSA, it is known that therapies are recommended according to the severity of the AHI. It is also important to emphasize the need for auxiliary measures in order to manage the sleep breathing disorder, such as weight loss; the elimination of sedatives, including tranquilizers, medication, and alcohol use; as well as positional therapies, when necessary[3].

CPAP has proved to be effective in severe cases by properly controlling systemic alterations[1]. However, when evaluating the use of CPAP in a prospective study of 121 patients, the initial adhesion (first three months) proved to be critical[5]. When Barnes et al. (2004)[18] analyzed and compared CPAP and OA adhesion, it was observed that 43% of the patients used CPAP for at least four hours per night in 70% of the nights, while 71% of them used AO for the same period, showing greater compliance to the oral devices[18]. Despite the lower adhesion to CPAP, it is still the gold standard treatment for OSA, as it has proven to be effective in controlling severe OSA. Therefore, OA is exclusively recommended in mild and moderate cases of OSA[3].

With respect to the OA, the literature has confirmed that mandibular advancement appliances may cause long-term side effects[11,18-22]. Despite the tooth movements that might occur during OA therapy[4-6,22], this therapy should not be discontinued unless the side effects prove to be greater than the benefits[2,11,13,19,23]. When analyzing lingual orthosis, that is, an OA with direct control over tongue, the concern regarding the side effects related to tooth movement is reduced.

A study conducted with tomographic analyses detected that both mandibular advancement devices and tongue-retaining devices showed a volumetric enlargement of the upper airway[14]. Direct lingual control was more effective than mandibular advancement in increasing the anteroposterior total area and volume. In severe and some moderate cases, mandibular advancement is unable to compensate for the obstruction caused by tongue collapse[11,13]. Therefore, lingual orthosis emerges as another treatment option for patients with mild and moderate OSA and as an alternative for those patients who refuse to comply with CPAP therapy. The present study also detected a decrease in central and mixed apneas. This reduction may have a correlation with the control of obstructive apneas, despite no scientific basis for such a correlation at this level.

According to the criteria of therapeutic success described in the literature[3,7,8], lingual orthosis presented an 18.1% success rate and was considered satisfactory in 81.8% of the

samples. Moreover, the lingual device also decreased the desaturation events and increased the minimum oxyhemoglobin saturation (SpO_2). However, it is important to show the possible side effects of lingual orthosis, such as difficulty swallowing, gagging, vomiting, and injury to the mucosa[17]. To prevent such effects, the lingual device must be carefully titrated, especially when it is necessary to place the spring in a position far below the oropharynx. An image (teleradiograph) to verify the spring positioning is also of paramount importance.

Long-term monitoring of patients in this sample should be performed to verify patient adherence, as well as to check the respiratory and sleep parameters through polysomnography exams. Further studies are warranted to evaluate the efficacy of lingual orthosis. A randomized clinical trial is an excellent study design to test the device, since this type of study reduces the bias when testing new treatments.

CONCLUSION

In this study, the short-term analyses showed that lingual orthosis was effective in controlling severe OSA, demonstrating a significant improvement in respiratory parameters. We believe that further studies with more robust designs will provide the scientific evidence necessary to conclude that this novel design of oral appliance could be included in the technical therapeutic arsenal that deals with obstructive sleep apnea. Moreover, long-term analyses should be conducted in order to check patient adherence to the therapy.

ACKNOWLEDGEMENT

We thank our colleagues: Jorge Machado Caram, Marcelo M. Quintela, and Maria de Lourdes Rabelo Guimarães, who provided insight and expertise that greatly assisted in the research.

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Avaliação de um novo tipo de aparelho intraoral para o tratamento de casos graves de síndrome de apneia obstrutiva do sono: série de casos

Objetivo: Este estudo teve como objetivo avaliar os parâmetros respiratórios de pacientes diagnosticados com síndrome de apneia obstrutiva do sono (SAOS) de gravidade severa, tratados com um novo tipo de aparelho intraoral (AIO): uma órtese lingual.
Métodos: Foi realizado um estudo de série de casos com 11 pacientes que sofriam de SAOS grave, índice de apneia-hipopnéia (IAH) > 30 eventos/ h. Todos os pacientes foram submetidos a polissonografia (PSG) de noite inteira pré e pós-tratamento e foram diagnosticados por um médico com base nos índices da PSG. Todos os pacientes selecionados apresentaram história de não adesão para dispositivos de pressão positiva contínua nas vias aéreas.
Resultados: Antes do tratamento, o IAH médio da amostra foi de 65,9 ± 30,5. Depois de atingir a titulação ótima do AIO, a PSG mostrou um IAH médio de 10,1 ± 5,5 ($p < 0,001$). Houve uma redução significativa nos eventos obstrutivos que diminuíram de 60,0 ± 31,7 para 8,5 ± 3,8 eventos / hora ($p < 0,001$). A saturação mínima de oxihemoglobina mostrou melhora significativa após o tratamento, tendo sofrido um aumento de 74,3 ± 7,8 para 83,4 ± 4,9 ($p < 0,05$).
Conclusão: A órtese lingual foi efetiva no tratamento da SAOS grave, mostrando resultados satisfatórios para a amostra atual. São necessários mais estudos para ampliar o conhecimento sobre a eficácia deste AIO.
Descritores: Apneia obstrutiva do sono. Aparelhos ortopédicos. Relatos de casos.