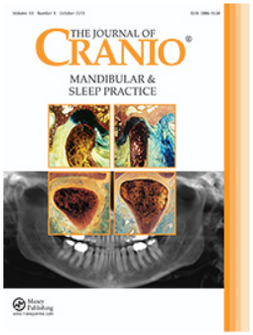

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CASE REPORT

Difficult-to-treat OSAS: Combined continuous positive airway pressure (CPAP) and mandibular advancement devices (MADs) therapy. A case report

Valeria Luzzi DDS, PhD[a], Marco Brunori MD, DDS[b], Sergio Terranova MD, DDS[c], Carlo Di Paolo MD, DDS[a], Gaetano Ierardo DDS, PhD[a], Iole Vozza DDS, PhD [a] and Antonella Polimeni MD, DDS [a]

[a]Department of Oral and Maxillo-facial Sciences, Head and Neck Integrated Activities Department, Policlinico "Umberto I", Sapienza University of Rome, Rome, Italy; [b]Respiratory Pathophysiology and Rehabilitation Unit, Policlinico Umberto I, Rome, Italy; [c]Private Practice, Rome, Italy

ABSTRACT

Background: Obstructive sleep apnea syndrome and is characterized by recurrent episodes of partial or complete upper airway collapse during sleep with consequent oxygen desaturations and cardiovascular, neurological, and metabolic impairment.

Clinical presentation: The authors report the case of a 66-year-old male presenting "metabolic syndrome" (obesity, impaired glucose tolerance, dyslipidemia, multi-drug treated arterial hypertension), atopy, mouth breathing due to turbinate hypertrophy, and pathological daytime sleepiness. As patient's compliance to standard continuous positive airway pressure (CPAP) therapy was poor, he was treated using low-pressure CPAP combined with a mandibular advancement device (MAD).

Conclusion: In selected patients, a treatment combining CPAP and MAD might be a more tolerable alternative to CPAP alone. The improved pharyngeal patency, promoted by mandibular advancement and stretching of the pharyngeal muscles, allows operating the CPAP at lower pressures when the MAD alone is not sufficient to induce a safe sleep profile.

KEYWORDS

Obstructive sleep apnea; airway pressure; mandibular advancement

Introduction

Obstructive sleep apnea syndrome (OSAS) belongs to the wide spectrum of sleep-disordered breathing [1–6] and is characterized by recurrent episodes of partial or complete upper airway collapse during sleep with consequent oxygen desaturations and cardiovascular, neurological, and metabolic impairment. The role of dentistry in its recognition and treatment is expanding. Evidence-based recommendations on therapeutic options in patients affected by severe OSAS indicate that continuous positive airway pressure (CPAP), administered by mechanical airway support through a nasal or facial mask, is the first-line choice, inducing a prompt restoration of the upper airways patency. Unfortunately, a large percentage of patients refuse or discontinue CPAP or ventilatory treatment, with serious clinical consequences. This is mainly due to claustrophobia and intolerance to the high CPAP pressures necessary for preventing collapse of upper airways.

Alternative therapeutic options include mandibular advancement devices (MADs), which are better tolerated than CPAP and show good compliance in real-life studies [7–9]. The overall efficacy of MADs achieves a 50% reduction of the apnea–hypopnea index (AHI) with a parallel reduction in snoring, daytime sleepiness, neuropsychological dysfunctions, and cardiovascular risks [7–12]. Side effects such as toothache and jaw pain are mostly mild. Short-term compliance is reported to be very high, while its 50% long-term (>2 years) compliance is higher than that of CPAP [13,14]. However, guidelines prescribe oral appliances in moderate OSAS without respiratory failure only as a second-choice option for patients who refuse CPAP. Recent research [15–17] suggests that the combined use of CPAP and MAD in a particular subset of OSAS patients could represent an additional option, since the improvement of pharyngeal patency, secondary to mandibular advancement, might allow the use of lower pressures of CPAP, resulting in better compliance to the treatment, as high pressure and airflow represent a major cause for CPAP discontinuation.

CONTACT Iole Vozza iole.vozza@uniroma1.it Department of Oral and Maxillo-facial Sciences, Sapienza University of Rome, Via Caserta 6-00161, Rome, Italy

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Case report

A 66-year-old Caucasian male patient presenting with "metabolic syndrome" (obesity with body mass index (BMI) of 31 kg/m^2, impaired glucose tolerance/insulin resistance, dyslipidemia, multi-drug-treated arterial hypertension), with a history of repeated episodes of seasonal and dust-related allergies, mouth breathing due to turbinate hypertrophy and reduced transversal diameters of the nasal cavities, and pathological daytime sleepiness, was referred in 2009 to the Department of Cardiovascular and Respiratory Diseases of Policlinico "Umberto I" in Rome for cardiorespiratory overnight monitoring. His daytime lung function tests showed normal spirometric values, while his night AHI was in excess of 40 events per hour, with no substantial differences between supine and right/left lateral positions, and his oxyhemoglobin desaturation index (ODI) was 24 events per hour, with a good average saturation, suggesting severe OSAS with irrelevant postural changes and severe snoring.

Together with weight loss and an ear, nose, throat (ENT) examination, after proper titration, a nocturnal CPAP therapy with 12 cm H_2O operating pressure was prescribed. Unfortunately, despite several trials with different nasal and oral masks, the patient did not tolerate this therapy, showing noncompliance to CPAP due to claustrophobia and excessive high pressures. Besides nasal steroid topical treatment, ENT examination indicated uvulopalatoplasty (UPP) as a possible surgical approach to OSAS. The patient agreed to the UPP intervention with the intent to evade CPAP treatment. The patient signed the informed consent, and the protocol used complied with the ethical guidelines of the 1975 Helsinki Declaration.

Two years after palate surgery, the patient returned to the clinic with persistence and aggravation of daytime sleepiness, poor control of blood pressure, despite appropriate medication, and BMI increased to 32 kg/m^2. A new cardiorespiratory examination confirmed the persistence of severe OSAS and, once again, nocturnal CPAP treatment was prescribed. Despite repeated titration sessions, using CPAP, autoCPAP, and Bi-Level modalities, several attempts with nasal pillows and silicone-gel oro-nasal masks, and pharmacological treatment of sleep fragmentation with Zolpidem, the patient still did not tolerate the treatment, mainly due to the high pressures required, and showed no compliance.

At the Head and Neck Integrated Activities Department of Policlinico "Umberto I" in Rome, the patient underwent a careful clinical evaluation (Figure 1). Extraoral examination showed a concave class III profile, while the intraoral clinical exam showed an occlusion with anterior and latero-posterior crossbite, due to sagittal and transversal hypodevelopment of the upper maxilla, and macroglossia. The latter, together with reduced intermaxillary space, forced the tongue in a high and retruded position within the oral cavity and gave it a scalloped aspect. This exam suggested, as an alternative therapeutic option, a maxillofacial surgical intervention for the correction of macroglossia with advancement and stabilization of the hyoid bone, rapid maxillary expansion, and bimaxillary advancement. The patient refused to undergo maxillofacial surgery.

At the same time, further sleep studies showed persistence and worsening of pathological AHI and ODI (AHI = 60.7, ODI = 40), confirming once again a very severe OSAS. After obtaining informed consent, an off-label mandibular advancement treatment was thus started using the Somnodent® oral appliance (Somnomed® Ltd.,

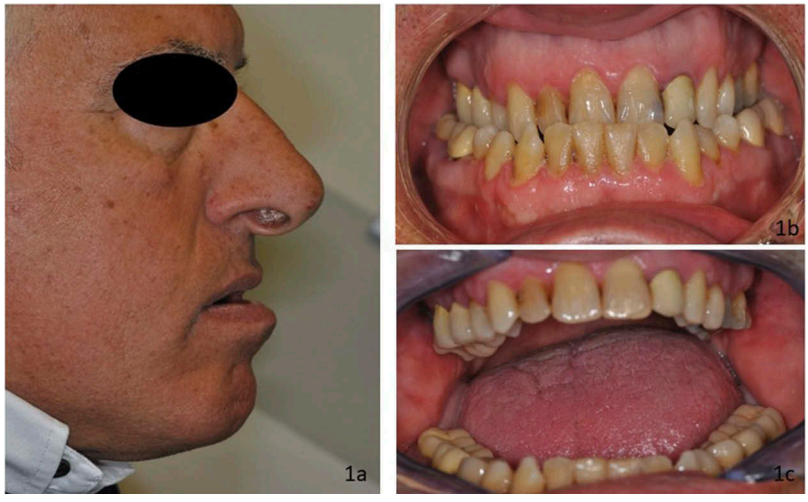

Figure 1. Extraoral examination shows a concave class III profile (a) while the intraoral clinical exam shows an occlusion with anterior and latero-posterior crossbite (b) and macroglossia (c).

Australia) (Figure 2) [18], followed by polysomnographic controls. The initial titration of the MAD was at 50% of maximum protrusion with progressive activations (1 mm per month) up to a total of 3 mm (Figure 3). The three-month follow-up showed an improvement from severe to moderate AHI and a good compliance to the MAD appliance. Follow-up visits did not show any long-term modification to the patient's bite.

However, although the ODI was reduced by half, the non-reassuring reduction of only 15 units of the AHI indicated a low effectiveness of the MAD treatment alone and recommended the reevaluation of the CPAP treatment. The new application, following a proper titration of the CPAP with a pressure of 12 cm H_2O, lowered the AHI to 11 and the ODI to 16.8. Again, the patient's noncompliance to CPAP led to a new discontinuation of the treatment.

Following a reevaluation of the case, to improve patient compliance, a new therapeutic strategy was chosen using a combined therapy with oro-nasal mask CPAP and MAD. The MAD guaranteed an increase of the vital oral airspace through the induction of postural and functional changes of the jaw skeletal tissues and of the lingual soft tissues, via a mandibular advancement and a front repositioning of the tongue, respectively. Thanks to the increased patency of the upper airways, these changes allowed the use of significantly lower CPAP operating pressures (6–7 cm H_2O, compared to 12 cm H_2O needed with the application of CPAP alone), as confirmed by a new polysomnography, run during combined MAD–

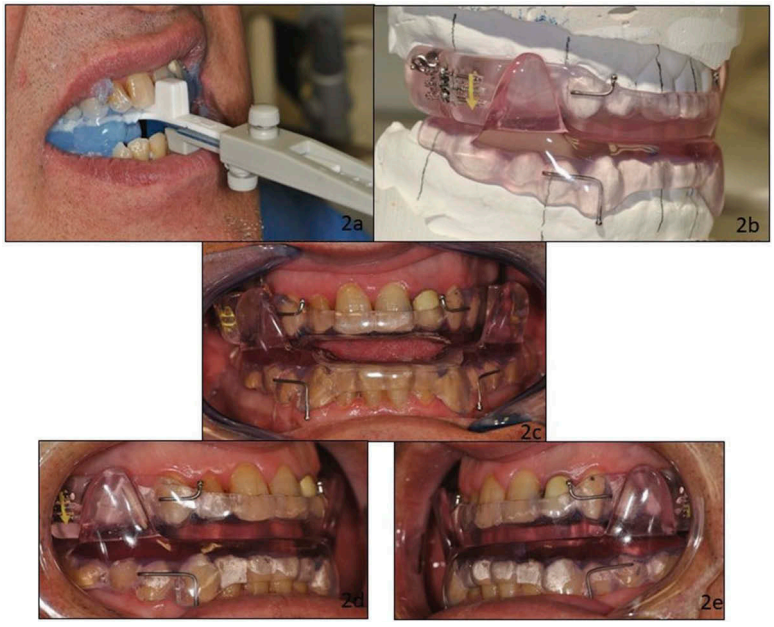

Figure 2. Preliminary intraoral impression measurement (a), adaptation of the Somnodent® MAD (b), and initial placement of the device: front (c), left (d), and right (e) views.

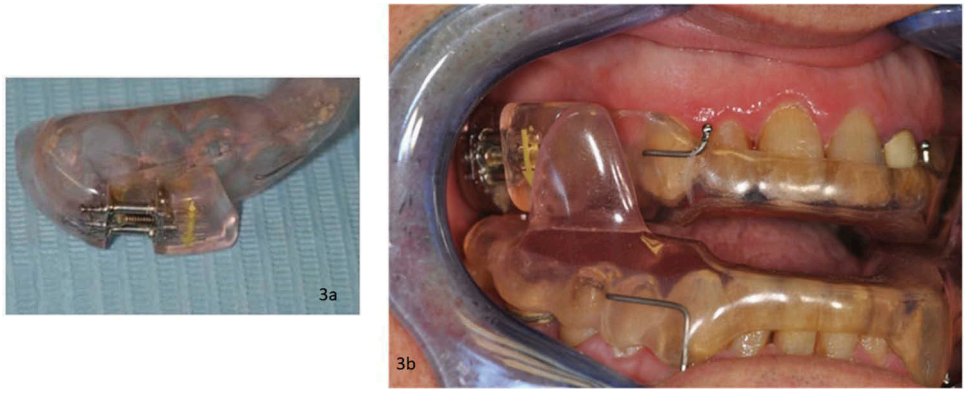

Figure 3. Detail of the MAD after activation (a) and final placement of the device (b).

Table 1. AHI and ODI values after each treatment stage.

Treatment stage	AHI	ODI
Before treatment	60.7	40
Treatment with MAD only	45	17.5
Treatment with CPAP to 12 cm H_2O	11	16.8
Combined treatment: MAD + CPAP to 7 cm H_2O	9.5	4.5

AHI: Apnea–hypopnea index; ODI: oxyhemoglobin desaturation index; MAD: mandibular advancement device; CPAP: continuous positive airway pressure.

CPAP therapy at low pressure, which showed a good control of apneic events and oxygen desaturations.

Table 1 summarizes the AHI and ODI values before treatment with only the application of MAD, with only the application of CPAP at a pressure of 12 cm H_2O, and with the combined application of MAD and CPAP at a pressure of 7 cm H_2O.

Discussion

The issue of compliance to the available OSAS treatments is the subject of debate in recent literature. A systematic review by Schwartz et al. [19] found a significantly higher compliance (+1.1 h per night) to MAD than to CPAP. This result is in disagreement with a previous systematic review by Li et al. [20], which found no statistical differences, a result confirmed by Sutherland et al. [21].

Clinical guidelines from the American Academy of Sleep Medicine[1] and the American Academy of Dental Sleep Medicine[2] recommend the use of a custom and titratable oral appliance, rather than no treatment, for patients suffering with OSAS who are not compliant with CPAP. The effectiveness of the treatment is additionally confirmed in a systematic review of the literature based on these guidelines [22].

This case report suggests that in difficult-to-treat OSAS with poor compliance to CPAP alone, due to side effects elicited by the elevated operating pressures, the combined use of CPAP and mandibular advancement might be an alternative option for selected patients, as also suggested in literature [23,24]. Pharyngeal patency, promoted by mandibular advancement and stretching of the pharyngeal muscles, might act as a tool to adopt lower operating pressures of the CPAP when MAD alone is not capable of reaching a safe sleep profile [18].

Conclusion

The authors reported a case of an OSAS patient, non-compliant to CPAP, who was successfully treated with a MAD in combination with low-pressure CPAP. Further studies are needed to evaluate the effectiveness of this combined therapy.

Disclosure Statement

The authors declare no conflict of interest.

ORCID

Iole Vozza DDS, PhD http://orcid.org/0000-0001-8545-9889
Antonella Polimeni MD, DDS http://orcid.org/0000-0002-2679-7607

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