Clinical management of a severe case of positional obstructive sleep apnea syndrome

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Abstract

Obstructive sleep apnea syndrome (OSAS) is a common disorder which involves upper airway collapse during sleep. Positional Obstructive Sleep Apnoea Syndrome is a sub-type of OSAS characterized by obstructive apneas mainly in the supine sleeping position. Mandibular advancement devices (MAD) are recommended as first-line therapy for mild to moderate OSA or as second-line treatment for severe OSA for patients who do not tolerate or respond to CPAP. A case report of severe positional OSAS (AHI/h 34.7) is presented. The clinical case was studied by CBCT head and neck scan and nocturnal polysomnography and treated with a mandibular advancement device, positional therapy, dietary adjustments and instructions for proper sleep hygiene. After the treatment the patient's AHI decreased to 5.1 and his general health improved. The MAD appliance represents a valid therapeutic alternative in subjects with severe OSAS who do not tolerate CPAP. A combined approach to the pathology can improve the patient's overall health and quality of life.

Keywords: OSA, Mandibular advancement devices, polysomnography, computed tomography.

Introduction

Obstructive sleep apnea syndrome (OSAS) is a common and treatable disorder, which involves upper airway collapse during sleep and results in intermittent hypoxaemia and sleep fragmentation (1,2). This disorder is the result of a complex interaction between anatomic factors, sleep-related factors, and central nervous system control over ventilation (3). The presence of some syndromes also predisposes the onset of sleep disorders (4). Brain dysfunctions might also manifest, such as excessive daytime sleepiness and lack of concentration and these are the most common causes of traffic accidents (5). The prevalence of OSA has been estimated to be 14% of men and 5% of women (6). Despite its prevalence, OSA is still an underdiagnosed medical condition, and more than 85% of patients with clinically significant OSA are never diagnosed (7). The presence of sleep disturbances is often associated with the presence of sleep bruxism (8). Positional Obstructive Sleep Apnoea Syndrome (POSAS) is a sub-type of OSAS in which obstructive apneas occur mainly in the supine sleeping position (more than 50% of apnea episodes). Polysomnography is the gold standard diagnostic test for the diagnosis of OSA in whom there is a concern for OSA based on a comprehensive sleep evaluation that include informations on sleep posture (9). The apnea-hypopnea index (AHI) is used to diagnose OSAS, and it is calculated as the number of apneas and hypopneas per hour during sleep (10). Mandibular advancement devices (MAD) are recommended as first-line therapy for mild to moderate OSA or as second-line treatment for severe OSA for patients who do not tolerate or respond to CPAP (11). Specifically, MADs interact with the mandible to the tongue, pharyngeal dilator muscles, and indirectly the soft palate. By moving the mandible forward, these structures that make up the lumen of the oropharynx are extended forward as well, thereby increasing the upper airway space (12). According to the literature Cone Beam Computed Tomography (CBCT) with its low effective radiation dose and low scanning time represents an effective technique for a 3D complete evaluation, when utilizing a large field of view protocol, for a comprehensive head and neck evaluation (13).

A case report of severe OSAS in a middle age man, studied by CBCT head and neck scan and nocturnal polysomnography and treated by customized and titrabled MAD, is presented.

Clinical case and therapeutic approach

A 41 years old Caucasian male, M.M., from the Department of Orthodontics referred by the Neurophysiopathology Department of Tor Vergata Hospital, with a chief complaint of snoring, numerous episodes of nocturnal apnea and daytime sleepiness. The patient had history of arterial hypertension and depression. The patient was undergoing pharmacological treatment for his depressive state. He had previously been treated for sleep apnea with CPAP. This treatment had not shown satis-

factory results due to the patient's poor tolerance to the therapy. The patient's body mass index was 27,13 (overweight) with a neck circumference of 45 cm, and he was being treated by a nutritionist to improve alimentation and lose weight. The patient was also instructed in proper sleep hygiene to improve his habits. The upper airway appeared normal in ear-nose-throat and laryngoscopy examinations. The patient's neck and tongue muscles were enlarged. Moreover the patient shows a reduced patency of the nasal airways passages due to hypertrophy of the nasal turbinates. The patient does not have any voluptuous habit. At the oral examination the patient showed right and left full class I with dentoalveolar biretrusion and increased OVB, signs of dental bruxism and normal-shaped arches. The patient besides shows macroglossia (Mallampati grade 3), normal size of the uvula and soft palate, type 1 tonsillar grading. Epworth sleepiness score (ESS) was 11. CBCT examinations was performed with NewTom VGi EVO unit (NewTom 3G, QR s.r.l.; AFP Imaging, Elmsford, NY, USA). The 3D reconstruction and evaluation of the upper airways showed an anatomical narrowing of the hypopharynx which predisposes the collapse of the airways during sleep. Written consent was obtained from the patient.

Upon polysomnographic examination, the subject was diagnosed with positional preference OSAS. During the test, he had 141 apneas, 1 of which were central. The patient's apnea hypopnea index (AHI) was 34.7, a value that meets the criterion for severe sleep apnea.

Because the patient refused CPAP therapy due to discomfort and considering the strong positional component of apneas, treatment options for reduction of the OSAS included a customized and titrabled MAD (Figure 1a) and a NS Sleep Positioner (Advanced Brain Monitoring, Carlsbad, CA) composed of a plastic device fastened on the back of the neck with an adjustable rubber strap secured by a magnetic clasp (Fig. 1b).

Customized MAD was obtained through the Gorge Gauge at maximum advancement without muscular pain for patients. The MAD chosen was the TELESCOPIC ADVANCER (Leone orthodontic products, Sesto Fiorentino, Firenze, Italy). Bimaxillary device with lateral connectors composed of a tube and piston mechanism with the possibility of adjusting the protrusion very accurately, with 7 mm of maximum elongation. Vestibular pin for elastic bands were added to the device to prevent the patient from keeping his mouth open while sleeping (Fig. 1a).

The MAD was periodically adjusted during monthly checks in order to obtain the maximum possible mandibular advancement in the absence of patient discomfort. After six months of using MAD and NS Sleep Positioner, the patient's apnea improved the patient reports a clear improvement in symptoms (ESS = 1), restful sleep, absence of asthenia upon awakening, absence of snoring and / or apneic episodes. The patient does not show any kind of discomfort, absence of signs or symptoms about temporomandibular dysfunctions. The second polysomnographic examination with MAD in situ revealed only nine apnea episodes and no one were central. The patient's AHI decreased to 5.1 (Fig. 2). Moreover, the patient's overall health improved owing to dietary adjustments. The patient appeared satisfied with the results of the therapy. A second TCCB with MAD in situ scan showed an increased dimension of the hypopharynx (Fig. 3).

Discussion

The patient was treated using a mandibular advancement device, positional therapy, dietary adjustments and instructions for proper sleep hygiene according with the existing evidenced-based clinical practice guidelines (14, 15). The patient presented in the basal PSG an AHI of 34.7 events/h. Before starting the therapy with the MAD, the patient tried CPAP for a few nights, once it is the primary treatment indication for severe sleep apnea. Nevertheless, he abandoned it as the discomfort was the no-compliance reason. After titrating the MAD, the results of the control polysomnographic, with the

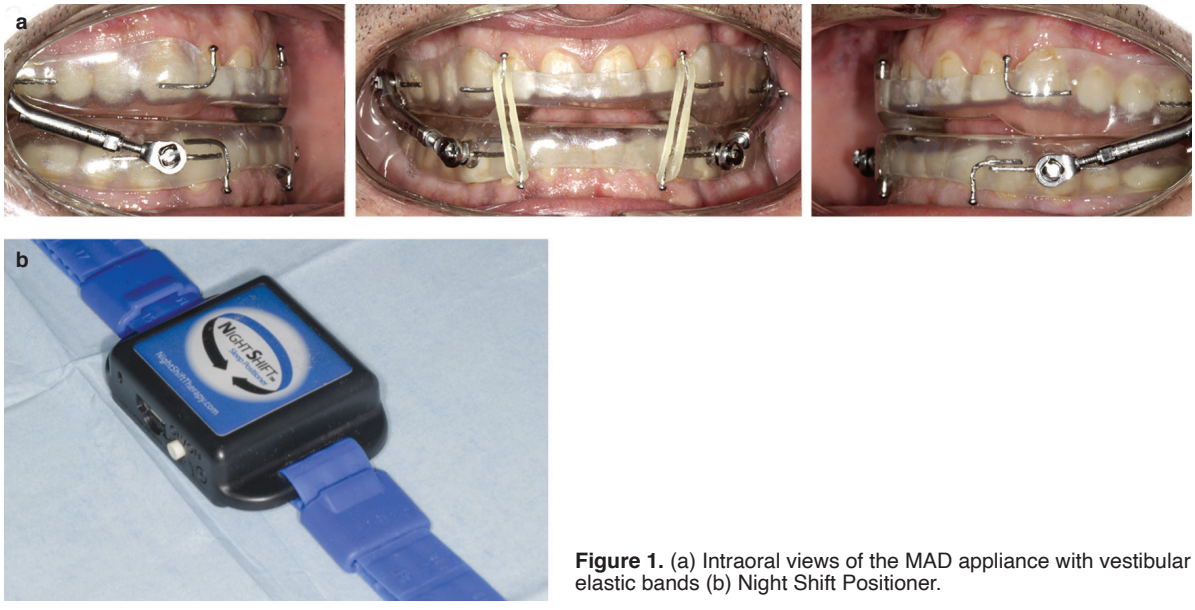

Figure 1. (a) Intraoral views of the MAD appliance with vestibular elastic bands (b) Night Shift Positioner.

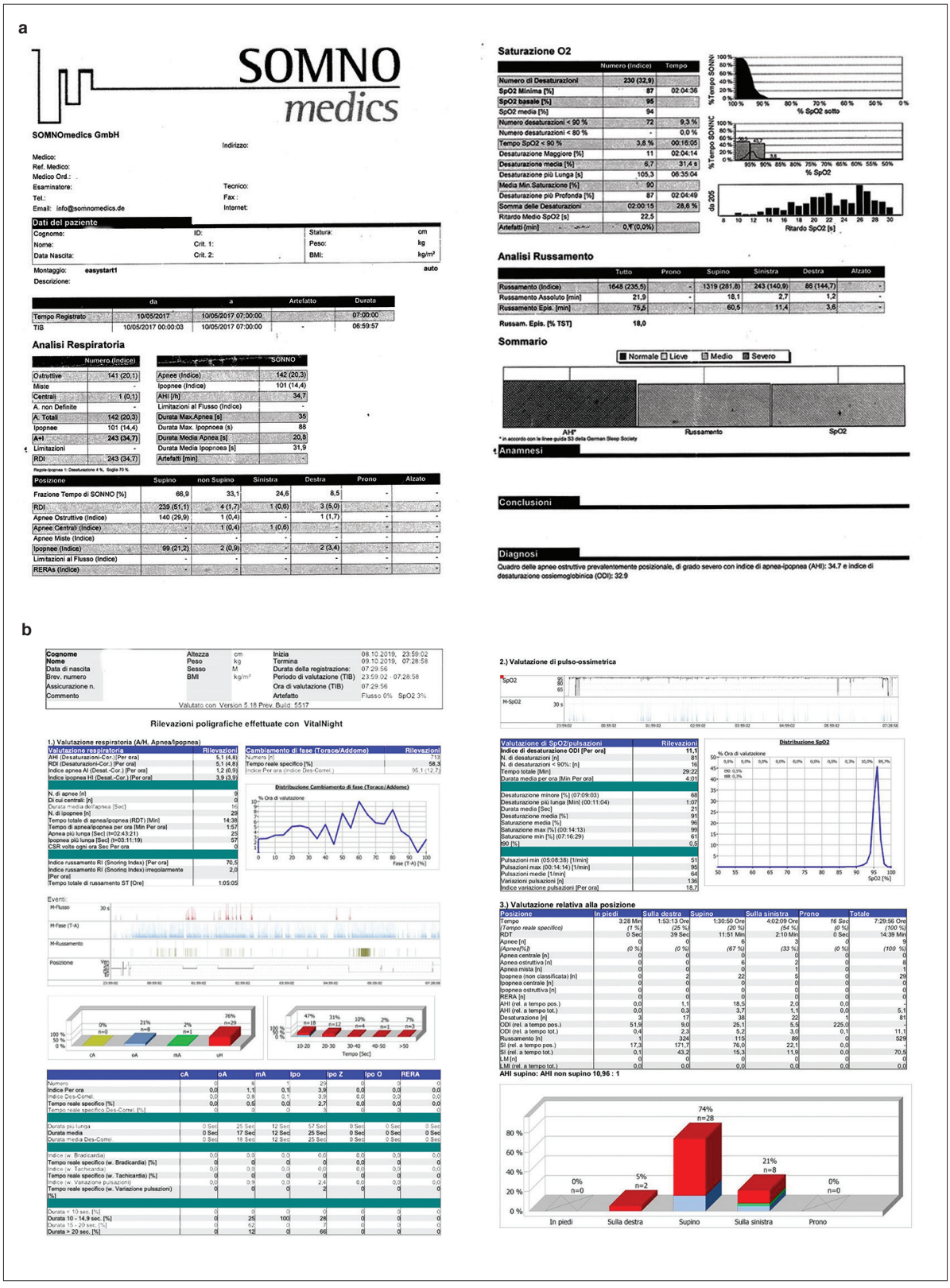

Figure 2. (a) Polysomnographic report before treatment (b) after treatment.

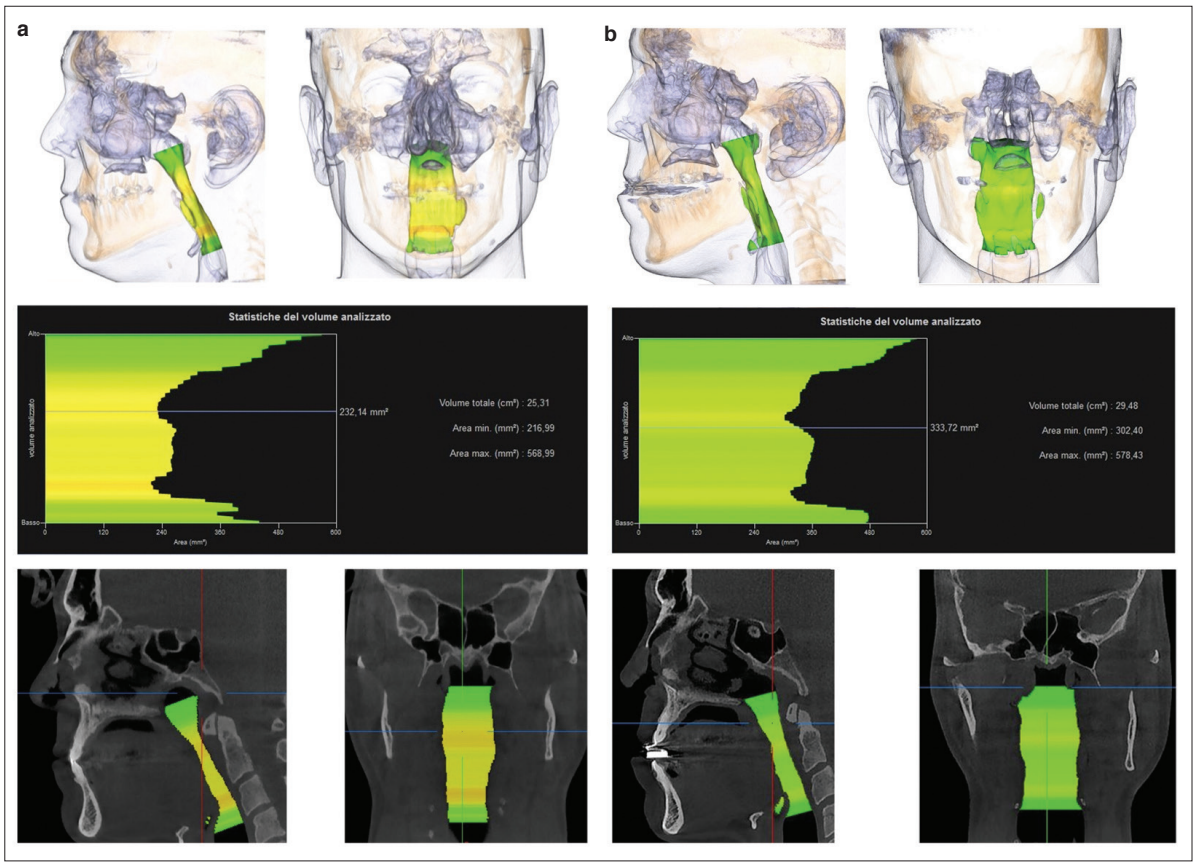

Figure 3. (a) 3D CBCT initial reconstruction and evaluation of the upper airways (b) with MAD in situ.

oral appliance in situ, showed an objective decrease in the rates of respiratory obstructive events. The full night PSG showed an AHI = 5.1 events/h. Patients with severe OSA have an increased cardiovascular risk, showing increased prevalence of hypertension, stroke, arrhythmia, and aortic events, although these findings are not always consistent (16-18). Without CPAP adherence, they must be treated with alternative therapies. As suggested in the literature, the efficacy of the OSAS therapy must be evaluated in relation to the patient's compliance (19). The main objective of MAD therapy is to reduce or normalize the AHI. There are some predictors that help dentists in the attempt to identify which patients will benefit more from the treatment. However, the predictors are still not fully clinically reliable (20). In the present case the patient was 41 years old, overweight and higher AHI. This outcome was not compatible with some strong predictors such as lower AHI, lower age, lower BMI and higher mandible protrusion (21). Despite this, the patient had an excellent response to MAD therapy.

In this study the patient wore a NS Sleep Positioner in order to avoid supine position. Van Kesteren et al. (22) demonstrated that, especially in POSA patients, the head position has a fundamental role in the genesis of apnoeas. Simply with head rotation, the anteroposterior collapse of the tongue due to gravity forces can be avoided and the airway can be stiffened. The body position is not only important to avoid the simple fall of the soft tissues by gravity but also for the effects on chest pressure. Joosten S.A. and colleagues (23) demonstrated a 13% functional residual capacity (FRC) decrease in the OSA group when moving from lateral to supine. This reduction of FRC influences upper airway collapsibility with resultant changes in upper airway pressure (24). The reduction in lung volume causes an increase in critical pressure (Pcrit) which influences the collapse of the upper airways causing the apnea event. Additionally, a reduced lung volume increases the ring gain of the ventilatory control system during reduced sleep lung oxygen and carbon dioxide deposits which can then function of respiratory instability in the supine position of sleep (25).

In this case report, the patient was placed on a weight-loss program with a nutritionist in order to manage the BMI. Although the effect of weight-loss on OSAS outcomes is not well studied, weight management is routinely recommended. A decrease in BMI is associated with improved metabolic outcomes in obese subjects and should be routinely encouraged notwithstanding is potential benefits regarding OSAS (26). Chirinos JA et al. (27) in his study has shown that CPAP, associated with a weight-loss program, increases insulin sensitivity and reduces serum triglyceride levels, but no improvement was observed with CPAP treatment alone, which

raises the important issue of the usefulness of adding weight-loss programs to CPAP treatment in order to improve the cardiovascular risk factor profile of obese patients with OSA. The same considerations can certainly also be made in the association between MAD and weight loss. A meta-analysis that included four randomized controlled trials assessing the effect of intensive lifestyle interventions on weight change and AHI reduction found that a weight loss of 14 kg was associated with a reduction in AHI of 16 events/h (28).

Conclusion

The mandibular advancement device associated with positional therapy, weight-loss program and instructions for proper sleep hygiene improved the polysomnographic parameters in a case of severe OSA. The MAD appliance represent a valid therapeutic alternative in subjects with severe OSAS who do not tolerate CPAP. A combined approach to the pathology can improve the patient's overall health and quality of life.

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