Successful Treatment of Extremely Severe Obstructive Sleep Apnea with a Dental Appliance

Yaron Haviv DMD PhD[1], Lilach Kemer MD[2], Roee Sheinfeld MD[3], Galit Almoznino DMD MSc MHA[1] and Gideon Bachar MD[4,5]

[1]Department of Oral Medicine, Hebrew University–Hadassah School of Dental Medicine, Jerusalem, Israel
[2]Sleep Medicine Institute, Assuta Medical Center, Tel Aviv, Israel
[3]Department of Neurology, Sheba Medical Center, Tel Hashomer, Israel
[4]Department of Otorhinolaryngology-Head and Neck Surgery, Rabin Medical Center (Beilinson Campus), Petah Tikva, Israel
[5]Sackler Faculty of Medicine, Tel Aviv University, Tel Aviv, Israel

ABSTRACT: **Background:** A dental appliance for obstructive sleep apnea (OSA) is recommended for patients who cannot adjust to continuous positive airway pressure (CPAP) treatments.

Objectives: To describe patients with extremely severe OSA who were successfully treated with a dental appliance and to compare their characteristics with the relevant literature to identify clinical features associated with a good outcome.

Methods: The clinical, management, and outcome data of three patients with an apnea-hypopnea index (AHI) of › 80 who showed clinical improvement following treatment with a dental appliance were collected retrospectively from sleep laboratory reports in Israel over a period of 3 years.

Results: The patients included one man and two women, aged 33, 56, and 61 years, respectively. The diagnosis of OSA was based on clinical examination and polysomnography. AHI values at presentation were 83, 81, and 84, respectively. Treatment with a dental appliance (Herbst® or MDSA®) was proposed due to patient noncompliance with CPAP. Follow-up polysomnography with the dental appliance revealed a reduction in the AHI to 1.7, 10.7, and 11, respectively. All patients had supine OSA and a retrognathic mandible, both of which have been found to be associated with a good prognosis for treatment with a dental appliance.

Conclusions: Dental appliances may be considered an appropriate second-choice option to treat severe OSA in patients who are noncompliant with CPAP. This study helps physicians identify patients with extremely severe OSA who are suitable for dental appliance treatment. Well-designed large-scale studies are needed to reach definitive conclusions.

IMAJ 2018; 20: 000–000

KEY WORDS: obstructive sleep apnea (OSA), dental appliance, continuous positive airway pressure (CPAP), apnea-hypopnea index (AHI), positional obstructive sleep apnea

Obstructive sleep apnea (OSA) syndrome is characterized by the frequent repetitive collapse and blockage of the upper airway during sleep [1]. Symptoms include snoring, choking, and gasping for air during sleep, daily sleepiness, and morning headaches. One of the main parameters used to assess the severity of the syndrome is the apnea-hypopnea index (AHI), which is calculated by summing the average number of apnea and hypopnea events per hour of sleep. An AHI of more than 30 is considered severe OSA by the American Sleep Disorder Association [2]. Severe OSA has been associated with cardiovascular morbidity [3], systemic hypertension, cerebrovascular disorders [4], reduction in overall quality of life [5], and a higher rate of fatal road accidents [6]. Race may be a factor in OSA severity [7]. Severe maternal OSA during pregnancy may affect neonatal neurological outcome, although mild maternal OSA apparently does not [8].

Continuous positive airway pressure (CPAP) is the first-line treatment for severe OSA [9]. However, its use is limited by poor patient adaptation and habituation [10], resulting in major health risks and necessitating other modes of therapy. Several studies have described the use of removable dental appliances that passively widen the upper airways by repositioning the mandible during sleep [Figure 1] [11].

Figure 1. Photograph of the Herbst® dental appliance for the treatment of obstructive sleep apnea

Although dental appliances yield poorer results than CPAP in terms of AHI reduction [12], they may be considered in patients who refuse or cannot tolerate CPAP. The 2006 guidelines of the American Academy of Sleep Medicine [13] recommended a dental appliance for snoring patients without OSA, patients with a minor to medium degree of OSA, and patients with severe OSA who cannot adapt to CPAP therapy. According to the 2015 updated guidelines, dental appliances are indicated for patients with primary snoring or adults with OSA who are intolerant of CPAP or prefer an alternative therapy regardless of the degree of OSA severity [14].

The aim of the present study was to describe patients with extremely severe OSA who were successfully treated with a dental appliance and to compare their characteristics with the relevant literature to identify clinical features associated with a good outcome.

PATIENTS AND METHODS

We describe three patients with extremely severe OSA (AHI > 80) who were successfully treated with a dental appliance at a tertiary medical center from 2014 to 2016. Successful treatment was defined as a reduction in AHI to the range of mild OSA or better. The clinical and outcome data of the patients were collected retrospectively from the sleep laboratory reports. The primary sleep tests were performed 1–3 years before presentation, and the subsequent sleep tests with the dental appliance were performed 6–8 months after onset of treatment.

Permission to publish facial photographs was provided by the patients [Figure 2, Figure 3].

PATIENT 1

A 33 year old man was referred by an ear, nose, and throat specialist. Primary complaints were snoring and daytime sleepiness. On physical examination, height measured 184 cm and weight 108 kg, for a body mass index (BMI) of 31.9. Past medical history revealed a diagnosis of severe OSA (AHI 84) at age 14 years, which was relieved subjectively by removal of the adenoids and tonsils. However, a home sleep test performed at age 31 years revealed an AHI of 83: supine AHI 91.5 (89.1% of the sleep time) and lateral AHI 13.3 (10.2% of the sleep time). Mean oxygen saturation was 93% (minimum 83%). Portion of the sleep test with < 90% oxygen saturation was 15.9%. Even after the patient was informed of the health risks of severe OSA, he refused CPAP. As a last resort, prior to bi-maxillary protrusion surgery, he opted to try a dental appliance. The patient was measured for a modified removable Herbst® mandibular repositioning device (Dentaurum GmbH, Ispringen, Germany). The Mallampati score was 1 [Figure 2]. Protrusive bite registration was performed with a raise of 0.9 cm. After the device was placed, the degree of mandibular advancement gradually increased to 12 mm beyond the regular bite, or approximately 90% of the maximum mandibular protrusive position. A follow-up portable home test with the dental appliance revealed a total AHI of 1.7 and an AHI of 2.1 in the supine position (81.9% of the sleep time). Mean oxygen saturation was 96% (minimum 91%), indicating no sleep apnea.

PATIENT 2

A 56 year old woman was referred by a sleep laboratory. Primary complaints were snoring and severe daytime sleepiness. On physical examination, height measured 158 cm and weight 89 kg, for a body mass index of 35.6. The patient was under treatment with apixaban (Eliquis®) for atrial fibrillation. On full polysomnography, AHI was 81 with minimum oxygen saturation at 74%. Oxygen saturation was below 90% for 23.2% of the sleep time. The oxygen desaturation index (ODI), defined as the number of times per hour of sleep the blood oxygen level drops by a certain degree, was 59.2. Supine AHI was 104.5 (52.3% of the sleep time) and lateral AHI was 13.3. The patient

Figure 2. Patient 1 (Herbst dental appliance), notable clinical features. Concave face, Mallampati 1, raised protrusive bite registration 0.9 cm with mandibular advancement of 12 mm

Figure 3. Patient 2 (MDSA dental appliance), notable clinical features: concave face, Mallampati 2, raised protrusive bite registration 0.9 cm with mandibular advancement 7 mm

was unable to adapt to CPAP or to lose weight. Therefore, an MDSA® device (MDSA, Middle Park, Victoria, Australia) was suggested. The Mallampati score was 2 [Figure 3]. Protrusive bite registration was performed with a raise of 0.9 cm. When the device was placed, mandibular advancement increased to 7 mm beyond the regular bite or approximately 70% of the maximum mandibular protrusive position. A follow-up full polysomnography with the dental appliance yielded an AHI of 10.7. Supine AHI was 15.3 (51.5% of the sleep time), and lateral AHI was 5.7. Minimum oxygen saturation was 84%; oxygen saturation was below 90% for only 3.4% of the sleep time, with an ODI of 5.8%. These findings indicated mild OSA. The patient reported significant alleviation of her nocturnal complaints and resolution of the daytime sleepiness.

PATIENT 3

A 61 year old woman was referred by a sleep laboratory. Primary complaints were snoring and frequent waking during the night to urinate. Height measured 153 cm and weight 65 kg, for a BMI of 27.8. A home portable sleep test revealed severe sleep apnea, with AHI 84 and minimum oxygen saturation 73%. A Herbst® mandibular repositioning device was suggested. Mallampati score was 2–3. Protrusive bite registration was performed with a raise of 0.9 cm. When the device was placed, mandibular advancement increased to 7 mm beyond the regular bite, or 80% of the maximum protrusive position. On the follow-up home sleep, AHI was 11 and minimum saturation index was 84%, indicating milder and less hazardous OSA.

RESULTS

The patients included one man and two women. The diagnosis of OSA was based on clinical examination and polysomnography. AHI values at presentation were 83, 81, and 84, respectively. Treatment with a dental appliance was proposed due to patient noncompliance with CPAP. Protrusive bite registration was performed with a raise of 0.9 cm. When the device was placed, mandibular advancement increased to 90%, 70%, and 80%, respectively, of the maximum protrusive position. Follow-up polysomnography with the dental appliance revealed a reduction in the AHI to 1.7, 10.7, and 11, respectively. All patients had supine OSA and a retrognathic mandible, both of which have been found to be associated with a good prognosis for treatment with a dental appliance. Characteristics of patients and dental appliances as well as results on sequential sleep tests are summarized in Table 1.

Table 1. Summary of patient characteristics and treatment outcome

	Patient 1	Patient 2	Patient 3
Gender	Male	Female	Female
Age, year	33	56	61
BMI, kg/m^2	31.9	35.6	27.8
Primary AHI	83	81	84
Supine AHI	91.5	104.5	–
Non-supine AHI	13.3	13.3	–
Minimum O2 level	83%	74%	73%
O2 < 90% (% of night)	15.9%	23.2%	–
Mallampati classification	1	2	2–3
Retrognathic mandible	✓	✓	✓
Appliance type	Herbst®	MDSA®	Herbst®
Vertical opening (cm)	0.9	0.9	0.9
Mandibular advancement*	90%	70%	80%
Final AHI	1.7	10.7	11
Supine final AHI	2.1	15.3	–
Non-supine final AHI	< 1	5.7	–
Final O2 < 90% (% of night)	0%	3.4%	–
Final minimum O2 level	96%	84%	84%

*Degree of mandibular advancement from maximum protrusion (%)
AHI = apnea-hypopnea index, BMI = body mass index, O2 = oxygen saturation

DISCUSSION

We describe a series of patients in whom dental appliances proved effective in reducing extremely severe OSA. Final AHI values were 1.7, 10.7, and 11. The strictest definition of successful treatment of OSA in the literature is a final AHI of less than 5 [15], which is considered normal breathing by the American Sleep Disorder Association. Although treatment with a dental appliance is well accepted for mild and moderate OSA [13], in patients with severe OSA, the reported success rates were relatively low. A recent study demonstrated a reduction in AHI from an average of 57 to 17 [16], but none of the patients had an AHI of more than 80 at outset.

Review of the relevant medical literature yielded several clinical factors that appear to play a contributory role in the success of dental appliance treatment for sleep apnea. The findings in the present series were analyzed against these data to identify clinical characteristics that could serve as good prognostic factors during the selection of candidates with extremely severe OSA for dental appliance treatment.

- Sleep position. Sleep position, supine or lateral, is well known to affect the successful use of dental appliances for OSA. Studies have suggested a possible association of sleep-dependent OSA with tongue base collapse/obstruction [17]. Accordingly, in our series, two of the three patients had supine sleep apnea (data were missing for the third patient), and the severity of the apnea decreased abruptly when they switched to side sleeping.
- Facial anatomic features. Retrognathic mandible and a concave profile have been reported to be positive predictors of good treatment outcome [18]. All of our patients had a concave profile [Figure 2, Figure 3]. We did not use cephalometric analysis because its predictive value is weak [19].

- Degree of mandibular advancement with the dental appliance. In general, the greater the degree of mandibular advancement with the device, the better the effect of treatment. However, potential side effects should be considered [18], especially in severe cases [20]. In our patients, 70% to 90% of the maximum mandibular protrusion was achieved with the device.
- Size of the vertical opening with the dental appliance. The value of this feature is controversial [21]. We think that in severe cases, it is better to use a larger vertical opening with consideration of the range of movement and side effects.
- Mallampati classification. The Mallampati classification is often used as a tool to predict the ease of intubation. A high score (class 3 or 4) is associated with a higher incidence of sleep apnea [22] and more difficult intubation. Our patients had relatively small tongues and short, soft palates, with Mallampati scores of 1 or 2. This may have influenced the results, although the Mallampati score has no known prognostic value for dental appliance therapy.
- Patient age. According to the literature, young patients seem to have a better chance of success with dental appliances [16]. One of our patients was 33 years old, but the two women were middle aged. Therefore, age was apparently not an important factor in our cases.
- BMI. A high BMI is a known risk factor in sleep apnea [23], but its effect on the success of dental appliance treatment is unclear [17]. Our patients had BMI values ranging from 27.7 to 35.6, which did not change over the course of treatment.

CONCLUSIONS

This study focused on the success of dental appliance treatment in patients with extremely severe OSA. On the basis of the literature, combined with the data derived from our extreme cases, we suggest that patients with supine OSA and a retrognathic mandible who are noncompliant with CPAP may have a relatively good chance of a positive response to dental appliance treatment.

This small study is intended to serve sleep laboratories, otolaryngologists, and dentists to more easily identify candidates with good prognostic features for dental appliance therapy. It does not discuss the use of such investigational tools as wake or sleep magnetic resonance imaging or other otolaryngology techniques, such as fiber optic laryngoscopy or drug-induced sleep endoscopy. Well-designed large-scale prospective studies are needed to better evaluate and quantify the factors associated with the success of dental appliance therapy in order to reach definitive conclusions.

Correspondence
Dr. Y. Haviv
Dept. of Oral Medicine, Hebrew University–Hadassah School of Dental Medicine, Jerusalem 91120, Israel
Phone: (972-2) 677-6151
email: yaronha@hadassah.org.il

References

1. Azagra-Calero E, Espinar-Escalona E, Barrera-Mora JM, Llamas-Carreras JM, Solano-Reina E. Obstructive sleep apnea syndrome (OSAS). Review of the literature. *Med Oral Patol Oral Cir Bucal* 2012; 17 (6): e925-9.
2. Sleep-related breathing disorders in adults: recommendations for syndrome definition and measurement techniques in clinical research. The Report of an American Academy of Sleep Medicine Task Force. *Sleep* 1999; 22 (5): 667-89.
3. Marin JM, Carrizo SJ, Vicente E, Agusti AG. Long-term cardiovascular outcomes in men with obstructive sleep apnoea-hypopnoea with or without treatment with continuous positive airway pressure: an observational study. *Lancet* 2005; 365 (9464): 1046-53.
4. Gibson GJ. Sleep disordered breathing and the outcome of stroke. *Thorax* 2004; 59 (5): 361-3.
5. Phillips CL, Grunstein RR, Darendeliler MA, et al. Health outcomes of continuous positive airway pressure versus oral appliance treatment for obstructive sleep apnea. *Am J Respir Crit Care Med* 2013; 187 (8): 879-87.
6. Horne JA, Reyner LA. Sleep related vehicle accidents. *BMJ* 1995; 310 (6979): 565-7.
7. Carel RS, Brodsky I, Pillar G. Obstructive sleep apnea: comparison of syndrome severity and risk factors for adult Jewish and Arab males in northern Israel. *IMAJ* 2015; 17 (8): 492-5.
8. Bassan H, Uliel-Sibony S, Katsav S, Farber M, Tauman R. Maternal sleep disordered breathing and neonatal outcome. *IMAJ* 2016; 18 (1): 45-8.
9. Sullivan CE, Issa FG, Berthon-Jones M, Eves L. Reversal of obstructive sleep apnoea by continuous positive airway pressure applied through the nares. *Lancet* 1981; 1 (8225): 862-5.
10. Kribbs NB, Pack AI, Kline LR, et al. Objective measurement of patterns of nasal CPAP use by patients with obstructive sleep apnea. *Am Rev Respir Dis* 1995; 147 (4): 887-95.
11. Ferguson KA, Cartwright R, Rogers R, Schmidt-Nowara W. Oral appliances for snoring and obstructive sleep apnea: a review. *Sleep* 2006; 29 (2): 244-62.
12. Lim J, Lasserson TJ, Fleetham J, Wright JJ. Oral appliances for obstructive sleep apnoea. *Cochrane Database Syst Rev* 2006:CD004435.
13. Kushida CA, Morgenthaler TI, Littner MR, et al. Practice parameters for the treatment of snoring and obstructive sleep apnea with oral appliances: an update for 2005. *Sleep* 2006; 29 (2): 240-3.
14. Ramar K, Dort LC, Katz SG, et al. Clinical practice guideline for the treatment of obstructive sleep apnea and snoring with oral appliance therapy: an update for 2015. *J Clin Sleep Med* 2015; 11 (7): 773-827.
15. Engleman HM, McDonald JP, Graham D, et al. Randomized crossover trial of two treatments for sleep apnea/hypopnea syndrome: continuous positive airway pressure and mandibular repositioning splint. *Am J Respir Crit Care Med* 2002; 166 (6): 855-9.
16. Haviv Y, Bachar G, Aframian DJ, Almoznino G, Michaeli E, Benoliel R. A 2-year mean follow-up of oral appliance therapy for severe obstructive sleep apnea: a cohort study. *Oral Dis* 2015; 21 (3): 386-92.
17. Marklund M, Stenlund H, Franklin KA. Mandibular advancement devices in 630 men and women with obstructive sleep apnea and snoring: tolerability and predictors of treatment success. *Chest* 2004; 125 (4): 1270-8.
18. Mehta A, Qian J, Petocz P, Darendeliler MA, Cistulli PA. A randomized, controlled study of a mandibular advancement splint for obstructive sleep apnea. *Am J Respir Crit Care Med* 2001; 163 (6): 1457-61.
19. Holty JE, Guilleminault C. Maxillomandibular advancement for the treatment of obstructive sleep apnea: a systematic review and meta-analysis. *Sleep Med Rev* 2010; 14 (5): 287-97.
20. Walker-Engstrom ML, Ringqvist I, Vestling O, Wilhelmsson B, Tegelberg A. A prospective randomized study comparing two different degrees of mandibular advancement with a dental appliance in treatment of severe obstructive sleep apnea. *Sleep Breath* 2003; 7 (3): 119-30.
21. Pitsis AJ, Darendeliler MA, Gotsopoulos H, Petocz P, Cistulli PA. Effect of vertical dimension on efficacy of oral appliance therapy in obstructive sleep apnea. *Am J Respir Crit Care Med* 2002; 166 (6): 860-4.
22. Nuckton TJ, Glidden DV, Browner WS, Claman DM. Physical examination: Mallampati score as an independent predictor of obstructive sleep apnea. *Sleep* 2006; 29 (7): 903-8.
23. Garg R, Singh A, Prasad R, Saheer S, Jabeed P, Verma R. A comparative study on the clinical and polysomnographic pattern of obstructive sleep apnea among obese and non-obese subjects. *Ann Thorac Med* 2012; 7 (1): 26-30.