

Oral Appliance Impact on Complex Sleep Apnea: A Case Report

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Complex sleep apnea is most often treated with adaptive servo-ventilation (ASV), a compensatory form of positive airway pressure therapy. What happens when ASV is not successful or tolerated? Is there a role for oral appliance therapy in the management of complex sleep apnea? A case is presented that explores these issues in a complex sleep apnea patient with comorbidities.

KEYWORDS: complex sleep apnea, oral appliance, mandibular advancement, sleep apnea, oral splint

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There is robust evidence supporting the use of mandibular advancement oral appliances in the treatment of obstructive sleep apnea for those patients preferring an oral appliance over continuous positive airway pressure (CPAP).¹ Some obstructive sleep apnea patients with no daytime hypercapnia are known to develop central apnea events during non-REM sleep, or a Cheyne-Stokes respiration pattern, with initial CPAP titration after resolution of airway obstruction events. This phenomenon has been termed complex sleep apnea. Some central events with initial CPAP titration may be transient and resolve with continued CPAP use. However, complex sleep apnea patients often have poor initial experiences with CPAP and become noncompliant. The cause of complex sleep apnea is currently unknown, although it may be associated with ventilatory control system instability (high loop gain). Though most commonly associated with the introduction of CPAP,² there are also case reports of treatment-related development of central sleep apnea following tracheostomy, maxillomandibular advancement surgery, and oral appliance therapy.^{3–6}

The prevalence of complex sleep apnea has been reported in a range of 1% to 20%.^{7,8} Risk factors may include congestive heart failure,⁹ though most patients exhibiting complex sleep apnea are not readily identifiable in advance. Successful treatment requires correction of both airway obstruction and ventilatory dysregulation. First-line treatment is adaptive servo-ventilation (ASV), a compensatory form of bilevel PAP that is less prone to precipitating central apneas. Not all patients are responsive to ASV, and alternative avenues for treatment are needed. There are few reports in the literature regarding the use of mandibular advancement oral appliances in the treatment of complex sleep apnea. The following is a case involving just such a treatment.

REPORT OF CASE

A 77-year-old Caucasian male was referred for consideration of oral appliance therapy by a board-certified sleep physician/pulmonologist. His body mass index was 30 kg/m² and his neck size was 16 inches at initial presentation. He expressed concerns about snoring, difficulty maintaining sleep, nightmares, night thrashing, and feeling tired all the time. His Epworth Sleepiness Scale score was 20. The relevant medical history included high blood pressure, Barrett's esophagitis, arthritis, and recurrent liposarcoma. Medications included metoprolol, valsartan, amlodipine, duloxetine, omeprazole, allopurinol, fentanyl

transdermal patch, and supplemental oxygen. The patient had completed a baseline split polysomnogram at an American Academy of Sleep Medicine accredited sleep center and received a diagnosis of severe obstructive sleep apnea with an apnea hypoxia index (AHI) of 69.8/h and moderate oxyhemoglobin desaturations to a nadir of 80% and T90 of 38.8%. During the CPAP titration portion of the study, central apneas were exchanged for obstructive apneas and a diagnosis was made of severe complex sleep apnea. He subsequently completed a bilevel PAP titration polysomnogram, but central apneas developed with increasing pressures and bilevel PAP treatment was not successful. He next received an ASV titration polysomnogram, but severe mask leaks were noted at all ASV pressures despite a trial of multiple interfaces and use of a chin strap. Treatment with ASV was not successful. It was at this point that the patient was referred for consideration of oral appliance therapy.

The patient had received routine dental work including a number of stable fillings and crowns. His overbite was 9 mm and overjet was 4 mm. There were no diastemas or abfractions, though moderate occlusal wear was noted. He exhibited moderate periodontitis including mild-moderate mobility of the incisor teeth. His skeletal profile was retrognathic (Class II), and occlusal classification was Class II Division 2 on the right and Class I on the left. The oropharynx was characterized by a very low-arch palate (Mallampati IV). He had a large tongue with scalloping of the lateral borders. The temporomandibular joints, muscles of mastication, and mandibular range of motion were within normal limits. He was deemed a suitable candidate for treatment with a mandibular advancement oral appliance and was encouraged to improve his oral hygiene as well as work with his general dentist to improve his periodontal health. The selected oral appliance was a TAP 3 because of the patient's compromised manual dexterity, as gradual home mandibular advancement adjustments are easily accomplished by the patient with this design. In addition, the TAP 3 design restricts the degree of mandibular opening, reducing the risk of further airway obstruction during wide opening.

The patient made steady progress with gradual home mandibular advancement calibration of the oral appliance. However, he reached a point where placement of the oral appliance became a challenge for him. He was instructed to reverse several appliance adjustments prior to seating the device and then readjust the appliance to the treatment position after placement. He was

unable to comply due to difficulty placing the adjustment key into the device after placement. The adjustment key was then permanently attached to the oral appliance with cyanoacrylate adhesive. This allowed the patient to comfortably follow through with the recommended protocol and the oral appliance calibration continued until reaching the patient's comfortable limit. His wife reported snoring was well controlled at this setting. However, the patient described only mild improvement in sleep continuity and significant ongoing daytime sleepiness. The patient consulted with board-certified sleep physicians at several major area accredited sleep centers before presenting for a split-night polysomnogram, with the first half of the night dedicated to ASV titration and the balance for TAP 3 oral appliance confirmation/calibration.

With ASV, the patient's AHI was 60.8 disordered breathing events (DBE) per hour which included a mixture of central and obstructive events. His blood oxygen desaturation nadir was 83%. Once again, ASV was unsuccessful. During the oral appliance portion of the test, the AHI was 6.9/h at the most optimal oral appliance setting (1 mm of additional mandibular advancement), and blood oxygen desaturation nadir was 96%. The interpreting physician made a recommendation of home therapy with use of the oral appliance and supplemental oxygen. The patient found his sleep continuity was improved. However, he continued to complain of nightmares, memory loss, and sleeping most of the day. Several months later, he completed a new oral appliance confirmation/calibration polysomnography (PSG) which found an AHI of 45/h and blood oxygen desaturation nadir of 87%. It was thought that a combination of oral appliance therapy with ASV might be an option, so an additional ASV titration PSG in the presence of the oral appliance was completed. The test results included an AHI of 58.8/h and blood oxygen desaturation nadir of 89%. There were minimal central events this time and the desaturation nadir was much improved compared to the baseline polysomnogram.

At the patient's next visit, he seemed much more alert and upbeat and said he was feeling better than he had in a long time. He described sleeping fairly well with the combination of his oral appliance simultaneous with ASV, and was feeling much more vigilant since his dose of fentanyl was recently reduced by a significant amount.

DISCUSSION

The clinical management of a challenging patient diagnosed with complex obstructive sleep apnea while being managed for chronic pain related to multiple surgeries for recurrent liposarcoma is described. This patient was resistant to established therapy for the complex sleep apnea condition. Oral appliance therapy was initially effective at improving polysomnographic variables when combined with supplemental oxygen. However, significant daytime symptoms remained and subsequent overnight sleep studies found treatment with oral appliance

therapy/O₂ to be subtherapeutic. Combination oral appliance therapy/ASV was considered to be the most stable treatment for this patient. Yet his sleep-related symptoms improved only after his dose of fentanyl transdermal was significantly reduced. Fentanyl is an opioid analgesic prescribed for the management of chronic pain.¹⁰ Known side effects include difficulty sleeping, daytime drowsiness, and respiratory depression. Side effects may be more pronounced in the elderly. It is not known if fentanyl may have contributed to objective signs of sleep disordered breathing, since no further sleep studies were obtained after the dose was reduced.

The care of this patient demonstrates a novel multidisciplinary approach to management of multifactorial symptomatology. This case demonstrates that oral appliance therapy has the potential to be both a precipitating factor in the development of complex sleep apnea and part of the solution. In addition, residual sleep-related symptoms invite further scrutiny of the patient's medical history and pharmacology. Comprehensive and coordinated care best enhances quality of life outcomes.

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