Case 18. "MAD That Stops Breathing"

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Saif Mashaqi and Taaha Rafi

History

This is a 67-year-old male with a past medical history of coronary artery disease, aortic stenosis, and high-degree atrioventricular (AV) block (status post pacemaker placement) who presented with complaints of excessive daytime sleepiness, vivid dreams, thrashing and kicking at night. His wife witnessed loud snoring but did not witness any apnea, gasping, or snorting. He denied any headaches in the morning. Sometimes, he woke up with dry mouth. His routine bedtime is around 9 PM and wake up time is 5 AM. He denied any difficulties falling asleep and mentioned two awakenings during the night. He takes two naps during the day (each for about 45–60 min). He endorsed symptoms of legs restlessness and urge to move them, which was worse at night, relieved by walking and exacerbated by rest.

Examination

The patient is awake, alert, oriented to person, place and time. He is not in distress, does not look sleepy or fatigued. Vital signs: HR 57, BP 154/90, SPO_2 99%, RR 8, body mass index (BMI) 27.1 kg/m^2. Examination of the head and neck did not reveal any nasal septal deviation, enlarged turbinate, or obstructed nasal flow. Mallampati score was II/IV. Tonsils were not enlarged (grade I). No retrognathia. No collapse of the ala nasi. No tenderness over the temporomandibular joint. Neck circumference is 15 in. (38.1 cm). The rest of the physical examination was within normal limits.

S. Mashaqi
Pulmonary, Allergy, Critical Care, and Sleep Medicine, Banner University Medical Center, University of Arizona College of Medicine, Tucson, AZ, USA
e-mail: saifmashaqi@email.arizona.edu

T. Rafi (✉)
Sleep Medicine, Banner University Medical Center, Tucson, AZ, USA

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Investigations/Studies

A diagnostic polysomnogram (PSG) was completed which showed mild obstructive sleep apnea (OSA) showing an apnea-hypopnea index—AHI 13.9/h. The study also showed excessive transient muscle activity during REM, and periodic limb movements of sleep (PLMS) index of 30.9/h.

Due to the patient's frequent travel to remote areas of the world, positive airway pressure (PAP) treatment was very inconvenient for him and he chose to pursue mandibular advancement device (MAD) therapy for the treatment of his mild OSA. Accordingly, he was referred to the department of sleep dentistry and was fitted with a MAD. Home sleep apnea testing (HSAT) was completed after the patient was fitted with the oral appliance. This study showed a respiratory event index (REI) of 16.4/h, mainly obstructive in nature. Clinically, the patient noticed improvement in his symptoms but due to his elevated REI he was referred to sleep dentistry for further advancement of the MAD. The device was advanced twice (over an 18-month period) followed by a full night PSG to evaluate the effectiveness of the MAD advancement. Clinically, he endorsed worsening of excessive daytime sleepiness and recurrent awakening although there was a minimal improvement in vivid dreams, thrashing and kicking at night. During the first 2 h of sleep the overall AHI was 88.9/h. There was a predominance of central sleep apnea with Cheyne-Stokes breathing (CSB) pattern, and the central AHI was 55/h. The study was then converted to a split-night polysomnogram, and the patient was treated with PAP therapy. At the conclusion of the study, the PAP mode in use was Bi-level PAP-ST (Table 18.1).

Table 18.1 Summary of sleep studies before and after MAD use

	Study 1 without MAD—baseline	**Study 2** with MAD	**Study 3** with maximum advancement of MAD
	(PSG)	(HSAT)	(PSG)
AHI (events/h)	14	16.4	89
OAI (events/h)	1.2	1.6	3.3
HI (events/h)	10	14.2	24
CAI (events/h)	2.3	0.6	55
m SpO_2 (%)	88	92	94
T-90 (%)	3.1	1	8

AHI = apnea-hypopnea index; OAI = obstructive-apnea index; HI = hypopnea index; CAI = central-apnea index; m SpO_2 = mean SpO_2; T-90 = % total sleep time with $SpO_2 < 90\%$; PSG = polysomnography; MAD = mandibular advancement device; HSAT = home sleep apnea test

Differential Diagnosis

Central sleep apnea secondary to MAD use.
Central sleep apnea with CSB
Central sleep apnea secondary to opioid use
Central sleep apnea secondary to medical condition
Idiopathic central sleep apnea

Discussion and Management

Our patient presented with a mild degree of obstructive sleep apnea, which was complicated by a severe degree of central sleep apnea with the use of MAD that was advanced twice. Clinically, the patient endorsed improvement in symptoms (witnessed snoring, apnea, excessive daytime sleepiness, recurrent awakening, vivid dreams, thrashing and kicking at night), especially in the early phase of treatment with the initial advancement of MAD. This was followed by worsening of these symptoms later with the maximal advancement of MAD.

The patient has history of aortic stenosis, and he underwent transcatheter aortic valve replacement. However, his ejection fraction on several echocardiograms was within normal limits making the diagnosis of central sleep apnea with Cheyne Stokes breathing secondary to heart failure with reduced ejection fraction (HFrEF) not possible. The patient does not have a history of stroke, neurological disorders, or end stage renal disease making the diagnosis of central sleep apnea secondary to medical condition less likely. He does not take opioids or narcotics for pain control. Since severe central sleep apnea developed in the context of using MAD (that was advanced twice) in association with worsening in symptoms, this makes this entity the most likely diagnosis. Treatment-emergent central sleep apnea associated with the use of MAD is not common and poorly understood. It was described in the literature of sleep medicine as early as 2006. Avidan et al. reported a case of mild OSA who elected to be treated with MAD. Although this helped with snoring, it did not help with excessive daytime sleepiness and follow up PSG showed new onset central sleep apnea (moderate), in addition to mild worsening in obstruction events [1]. This thought to be related to MAD. More recently, another case report was published by Mohan et al., presenting a patient with a moderate degree of OSA who was treated with MAD and developed new onset central sleep apnea with MAD (mild), which resolved after 1 year of acclimation [2]. Whether the pathophysiology of these central events is similar to complex sleep apnea (i.e., PAP treatment emergent central sleep apnea) is not fully understood.

Final Diagnosis or Most Likely Diagnosis

Central sleep apnea secondary to mandibular advancement device use.

References

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