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Case report

Sexsomnia and REM- predominant obstructive sleep apnea effectively treated with a mandibular advancement device☆

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ABSTRACT

Parasomnias with sexual behavior or sexsomnias are considered a subtype of NREM parasomnias. Obstructive sleep apnea/hypopnea (OSAH) has been described as a known triggering factor for parasomnias including sexsomnia. Nasal continuous positive airway pressure (nCPAP) has been the standard of treatment for OSAH but mandibular advancement devices (MAD) are becoming an important treatment alternative. We present the case of a patient with mild OSAH and sexsomnia who had resolution of both conditions with a MAD. This patient had the added uniqueness of having REM-predominant OSAH

1. Introduction

The third edition of the International Classification of Sleep Disorders lists parasomnia with sexual behavior, or sexsomnia, as a subtype of NREM parasomnias [1]. There have been approximately ten published cases that describe obstructive sleep apnea/hypopnea (OSAH) as a triggering factor for sexsomnia [2,3]; partial arousals have been described as the mediating factor. Treatment of OSAH with nasal continuous positive airway pressure (CPAP) was reported in five of the ten patients and was associated with resolution of the sexsomnia activity in all but one of the reported cases [2–4]. Mandibular advancement devices (MAD) are becoming an important therapeutic tools in the treatment of OSAH but its use has not been reported in patients with OSAH and parasomnias. We present the case of a patient with OSAH and sexsomnia who was fitted with a MAD and had resolution of both the OSAH and the sexsomnia episodes.

2. Presentation of the case

This is the case of a 27-year-old male who was originally referred to a specialized sleep medicine clinic for evaluation and treatment of possible sleep apnea and during the course of his evaluation reported abnormal sexual activities during sleep. The patient and his wife expressed concerns about him having sexual behaviors 1–2 times per month while he was asleep. The behaviors that were reported included attempts to initiate sexual intercourse and attempts to disrobe his wife at which time she would awaken him and this would cease his activity. The patient did not have recollection of the episode immediately after awakening. These episodes date back to three years, since the patient had been married, prior to presenting to the sleep clinic. He had a remote history of sleepwalking when he was a child that resolved spontaneously. He also had very vivid dreams whenever he slept for a long time or during daytime naps, some of the dreams had a sexual component. There was no report of any other dream enactment behavior. His sexual activity was reported as happening “in the middle of the night” but a more precise timing was not available, however, his dreams with sexual content were more frequent towards the end of the sleep period. The patient was a habitual snorer but his wife was unable to confirm or deny any relationship between the snoring and the sexual episodes or dreams.

The patient did not have any significant medical or psychiatric history. He had no family history for any parasomnia. He was not taking any medications at the time of his evaluation or the previous days. He did not have a history of illicit substance use and consumed a low amount of alcohol with less than 1 drink per day.

Epworth Sleepiness Scale (ESS) on presentation was 12/24.

Abbreviations: AHI, apnea/hypopnea index; CPAP, continuous positive airway pressure; ESS., Epworth Sleepiness Scale; MAD, mandibular advancement device; NREM, non-rapid eye movement; OSAH, obstructive sleep apnea/hypopnea; PSG, polysomnography; REM, rapid eye movement; vPSG, video polysomnography

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Table 1
Summary of PSG findings.

	TRT	TST	%N1	%N2	%N3	%REM	T -AHI	REM AHI	NREM AHI	Supine AHI	Non supine AHI	AI	Time < 90% O2
Baseline PSG	504 min	500	4.9	47.0	23.3	24.8	6.9	16.1	3.8	11.5	2	8.2	0.2 min
Post-Treatment vPSG	443.8 min	433.7	5.6	54.6	23.0	16.8	1.6	1.7	1.6	2.8	0.4	3.2	0 min

TRT: total recording time; TST: total sleep time; %N1: percentage of stage N1 sleep; %N2: percentage of stage N2 sleep, %N3: percentage of stage N3 sleep; %REM: percentage of stage REM sleep, T-AHI: total AHI; AI: arousal index, Time < 90% O2: cumulative time with an oxygen saturation below 90%.

3. Physical exam

Weight = 78 kg, body mass index = 23 kg/m^2

Oral and Dental exam: Class I occlusion, Mallampati class III, Friedman Tonsils I, no scalloped tongue, no temporo-mandibular joint crepitus, good oral health (no caries, good periodontal status).

Nasal: Good nasal patency, normal nasal breathing pattern.

Rest of the physical exam including a detailed neurologic exam was normal.

4. Diagnostic Polysomnography (PSG)

The patient underwent a diagnostic PSG in a specialized sleep laboratory. The PSG was scored by a polysomnography technician and reviewed by a sleep specialist. Respiratory events were scored using the American Academy of Sleep Medicine scoring manual with 3% oxygen desaturations for hypopneas.

Total sleep time was 481 min, sleep efficiency was 96%, arousal index was 8.2 events/hour.

There was evidence of mild OSAH with apnea-hypopnea index (AHI) of 6.9 events/hour, REM AHI was 16.1 events/hour, NREM AHI was 3.8 events/hour. This PSG study did not include video recording REM atonia was preserved. See Table 1 for more details.

5. Treatment

The patient was diagnosed with mild OSAH based on the results of the PSG as well as a diagnosis of parasomnia with sexual behaviors or sexsomnia based on the clinical history. The patient refused nCPAP and treatment was initiated with a RESMED Narval MAD® at 50% of maximal protrusion (7 of 14 mm). A dentist who specializes in sleep medicine fitted the MAD.

6. Post-treatment PSG

Post treatment video-PSG (vPSG) was completed using the same scoring rules as the diagnostic PSG and showed resolution of OSA as demonstrated by normalization of the AHI to 1.2 events/hour. This second PSG was completed with video recording and no parasomnia activity was observed; no abnormal arousals were observed from stage N3 sleep, REM atonia was also present during this second study. See Table 1 for more details.

7. Follow up

The patient had resolution of the sexual episodes, snoring, and excessive daytime sleepiness soon after treatment with the MAD was initiated; post treatment ESS was 8/24. The patient returned for follow up visits with his wife 3, 6, and 12 months after initiating treatment with MAD and he continued using the device every night with excellent tolerance.

8. Discussion

Mandibular advancement devices are becoming an important therapeutic tool in the treatment of OSAH. Our patient, like many other patients, did not want to pursue PAP treatment and the MAD provided an acceptable solution that worked very well. The results observed with the MAD in our patient are consistent with the results that were observed when nCPAP was used in other patients [2,3]. To our knowledge, this constitutes the first case described in the literature of a patient with sexsomnia and obstructive sleep apnea that are both effectively treated with a MAD. The exact mechanism connecting the parasomnia with sexual activity is unknown but we can hypothesize that frequent respiratory events led to abnormal arousals; it is unclear if his previous history of sleepwalking increased his risk for the parasomnia with sexual behavior. Treatment of underlying OSAH is recommended for almost all REM and NREM parasomnias but we were unable to find other published cases reporting the use of a MAD in a patient with OSAH and a parasomnia. Larger studies are necessary to gain a better understanding of the efficacy and safety of MADs in patients with parasomnias and OSAH. This case is also very interesting because the patient had REM predominant OSAH and while sexsomnia is considered a subtype of NREM parasomnias, this case adds to the possibility that sexsomnia could be related to both REM and NREM sleep.

Disclosure statement

This was not an industry-supported study. The authors have indicated no financial conflicts of interest. This manuscript does not cover the off-label use of any medication.

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