



Clinical letter

Control of focal impaired awareness seizures with an oral appliance in a patient with sleep apnea

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1. Introduction

Focal impaired awareness seizure (FIAS) is a clinical phenomenon with a prevalence of 0.8% that manifests as epileptic discharges commonly located in the amygdala-hippocampal complex. It can cause visual delusions and olfactory, auditory, or affective hallucinations [1]. Obstructive sleep apnea (OSA) is common in patients with epilepsy [2]. Despite their limitations, some studies have revealed an important relationship between OSA and refractory epileptic seizures (RES), as well as between OSA improvement and a corresponding RES improvement with adequate continuous positive airway pressure (CPAP) [3]. Mandibular advancement devices (MADs) have been proven to be effective in patients with OSA. Although typically contraindicated in seizure disorders, the efficacy of MADs in the treatment of OSA-associated epileptic events was reported by Fenton JE et al. [4] recently, suggesting its relevance in specific circumstances such as the comorbidity of OSA and epilepsy. In this report, we present a clinical case of a patient diagnosed with FIAS and moderate OSA who was treated with MADs.

2. Case report

A 49-year-old woman with a body mass index of 23.7, who is married and works for a marketing business, was referred by a neurologist after she was diagnosed with OSA, indicating her candidacy for treatment with MADs. During anamnesis, she reported a diagnosis of epilepsy, accompanied by bouts of focal impaired awareness occurring at least twice monthly, during which she would remain open-eyed and make repetitive movements with her hands and mouth; this was always followed by a strong migraine. Her first episode was at the age of 15. She used to be an athlete but had for a long time led a sedentary life. Her medications for epilepsy included carbamazepine (400 mg, 200 mg, and 400 mg; controlled-release) t.i.d.; and a sedative anxiolytic, clonazepam (10 mg), b.i.d. Despite taking her medication regularly, she had recurrent seizures on 10/30/14, 11/15/14, 11/25/14, and 12/28/14, with 1–3 daily bouts. She complained of interrupted and restricted sleep. Her previous medical history included a diagnosis of breast cancer at the age of 36 years with subsequent total mastectomies of

both breasts associated with early menopause. Additionally, her medical history was negative for smoking, alcohol consumption, use of illicit drugs, and other local/systemic pathological conditions. She presented as normotensive, with a mean blood pressure of 120/80 mmHg. Her usual sleep and waking times were around 11:00 p.m. and 6:30 a.m., respectively, but she would often wake up about three times in the night. However, she had no history of excessive daytime sleepiness (6/21 on the Epworth Sleepiness Scale). No dental or periodontal pathology was noted, and she presented with a normal hard and soft palate, long uvula, edentate tongue (with a Mallampati grade IV), and normal bite and occlusion. A cardiorespiratory study was indicated on 01/18/2015 due to a high probability of OSA (she exhibited respiratory pauses despite the absence of sleepiness or any reported snoring), revealing an apnea-hypopnea index (AHI) of 15 events per hour, without desaturation (Oxyhemoglobin Desaturation Index (ODI) = 2 events per hour (EV/H), and 5 snoring events. The patient refused ventilatory support with CPAP and treatment with an oral device was indicated. A MAD, SomnoDent, was manufactured with an initial advance of 7 mm. The patient's range of advancement was 13 mm, from –3 to +10. One week after the installation of the device she reported sleeping better without complaints of joint pain, and began an advancement of 8 mm. Given the persistence of slight snoring, witnessed by her husband, and the absence of side effects, an advance of 9 mm was initiated. Once clinical stability was confirmed, a control level III polysomnography was requested. After seven months, she reported a seizure, followed by a migraine, without any changes in sleep quality or snoring. The cardiorespiratory polygraph revealed an AHI of 7 EV/H, with 29 events of flow limitation; and ODI of 1 EV/H, with one event of snoring—subsequently, an advance of 10 mm was initiated for optimal therapy. During an appointment eight months later, she reported better sleep, no snoring, and had no complaints of headaches or seizures for four months. During a follow-up visit, on 05/05/17, it was noted that therapeutic stability was maintained without epileptic seizures since February 2016. On June 2017, a control level II sleep study showed a sleep duration of 374.5 min, an AHI of 3.4 EV/H, and an ODI of 1 EV/H, sporadic snoring, an arousal index of 6.1, and the absence of epileptiform activity or other changes.

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3. Discussion

In OSA patients, the number of epileptic events is related to intermittent hypoxia and the increased number of state transitions often observed during sleep. CPAP treatment of OSA in patients with epilepsy has been associated with improvement in seizure control [3]. Despite the fact that this patient was not diagnosed with a level I or II sleep study (integrating electroencephalogram) because of the urgency of a diagnosis for OSA, it is relevant to consider that sleep-related breathing disturbances, even without clinically significant desaturations, result in sleep instability leading to increased cortical activity, and therefore higher numbers of sleep transitions and subsequent seizure related events. Only one case reported in the literature showed improvement in epileptic events in a patient with OSA treated with a MAD [4]; although this case had a different type of the disease and a relatively restricted impact on patient outcome, it permitted the maintenance of combined CPAP and MAD therapy. In the present case using the MAD completely controlled the sleep apnea as well as the epileptic events, with a clear improvement on either subjective or objective sleep parameters.

4. Conclusion

This work corroborates previous theories on the therapeutic benefits

of MAD in patients with OSA and epilepsy. Furthermore, for the first time, the achievement of a full and rather stable therapeutic outcome with this therapeutic approach was demonstrated.

Conflict of interests

All authors declare no conflict of interests.

References

- [1] Fisher RS, Cross JH, French JA, et al. Operational classification of seizure types by the international league against epilepsy: position paper of the ILAE commission for classification and terminology. *Epilepsia* 2017;58(4):522–30.
- [2] Devinsky O, Ehrenberg B, Barthlen GM, Abramson HS, Luciano D. Epilepsy and sleep apnea syndrome. *Neurology* 1994;44(2060).
- [3] Vendrame M, Auerbach S, Loddenkemper T, Kothare S, Montouris G. Effect of continuous positive airway pressure treatment on seizure control in patients with obstructive sleep apnea and epilepsy. *Epilepsia* 2011;52:168–71.
- [4] Fenton JE, Fitzgerald C, Dillon PJ, O'Shea D. Never say never: circumventing a contraindication to control apnoea-induced epileptic events with a mandibular advancement device. *J Laryngol Otol* 2018;132(Nov. (11)):1036–8.