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Video-Clinical Corners

Comorbid episodes of primary bruxism and bruxism as an epileptic activity-related motor event

Jitka Bušková [a, b, *], Jana Košťálová [a], Eva Miletínová [a, b], Petr Bušek [c]

[a] National Institute of Mental Health, Klecany, Czech Republic
[b] Third Faculty of Medicine, Charles University, Prague, Czech Republic
[c] Department of Neurology, Centre of Clinical Neurosciences, First Faculty of Medicine, Charles University and General University Hospital, Prague, Czech Republic

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1. Introduction

Tooth grinding is typically observed as a main feature of benign sleep bruxism [1], which is a common sleep disorder with a prevalence of about 8% in the adult population [2,3]. Bruxism is typically associated with sleep arousal and is considered part of an arousal response [4]. We report a patient suffering from idiopathic generalized epilepsy (IGE), who presented both episodes of primary bruxism and bruxism related to epileptic activity. We believe that this case could contribute to better understanding of the relationship between sleep instability and epilepsy.

2. Case description

Our patient is a 24-year-old man previously diagnosed with anxiety disorder [5], currently complaining of long-lasting stress. The reason he came to our sleep laboratory was mainly his subjective difficulty in maintaining sleep, which was restless and nonrestorative, and tooth damage due to bruxism.

Previously, he was diagnosed with IGE with absences and generalized tonic-clonic seizures (GTCS) that have been present since adolescence (juvenile absence epilepsy). He was not fully compensated on levetiracetam, with the last GTCS seizure occurring one year prior to the examination at our laboratory, but with persisting absences. He was not aware of any parasomnia episodes. Our patient had been a heavy smoker since the age of 16, and during past three years he smoked 10 cigarettes per day. Currently, he drinks about one pint of beer daily and smokes marijuana every time he has difficulties falling asleep. He has a family history of bruxism.

Physical examination revealed severe tooth wear; no other pathological findings were reported. Our patient underwent one night of polysomnography (PSG) with 19-channel electroencephalography (EEG, International 20-10 Electrode System Placement), electrooculography, electromyography of mm. submentales and mm. tibiales ant., respiratory channels with electrocardiography and oximetry, and brain 3 T magnetic resonance imaging (MRI).

Video-PSG showed fragmented sleep with a Sleep Period Time of 412 min (SPT), Total Sleep Time 378 min (TST), sleep latency 28 min, REM sleep latency 163 min, Sleep Efficiency 91.7%, N1 stage 8.4% SPT, N2 stage 54.1% SPT, N3 stage 11.7% SPT, REM stage 17.5%, Wake after sleep onset 8.3%. In sleep stages N1-3, we detected episodes of sleep bruxism during the recording and epileptic discharges or subclinical electrographic seizures. The bruxism occurred isolated, without relation to epileptic discharges or bound to epileptic discharges (Fig. 1). The subclinical electrographic seizures characterized by generalized high-frequency EEG activity with bifrontal predilection of up to 5 s at the beginning of the seizure, followed by development of high-amplitude slow activity and SW (spike-wave) complexes lasting for 40–50 s alternate with episodes of bruxism (Video 1 and 2). The episodes of bruxism that occurred alongside the electrographic seizure several times lasted longer compared to those occurring out of the seizure [12.6 (range 8–27) vs. 4.8 (range 3–10) bursts], but otherwise their pattern was identical. The level of consciousness was not ascertained. No episodes of bruxism or epileptic discharges were detected in REM sleep. Brain MRI scans showed normal results. We concluded that in addition to IGE, our patient also suffered from anxiety disorder with comorbid insomnia, and bruxism.

Supplementary video related to this article can be found at https://doi.org/10.1016/j.sleep.2019.06.009.

* Corresponding author. Department of Sleep Medicine, National Institute of Mental Health, Topolová 748, 250 67 Klecany, Czech Republic.
E-mail address: vankjit@seznam.cz (J. Bušková).

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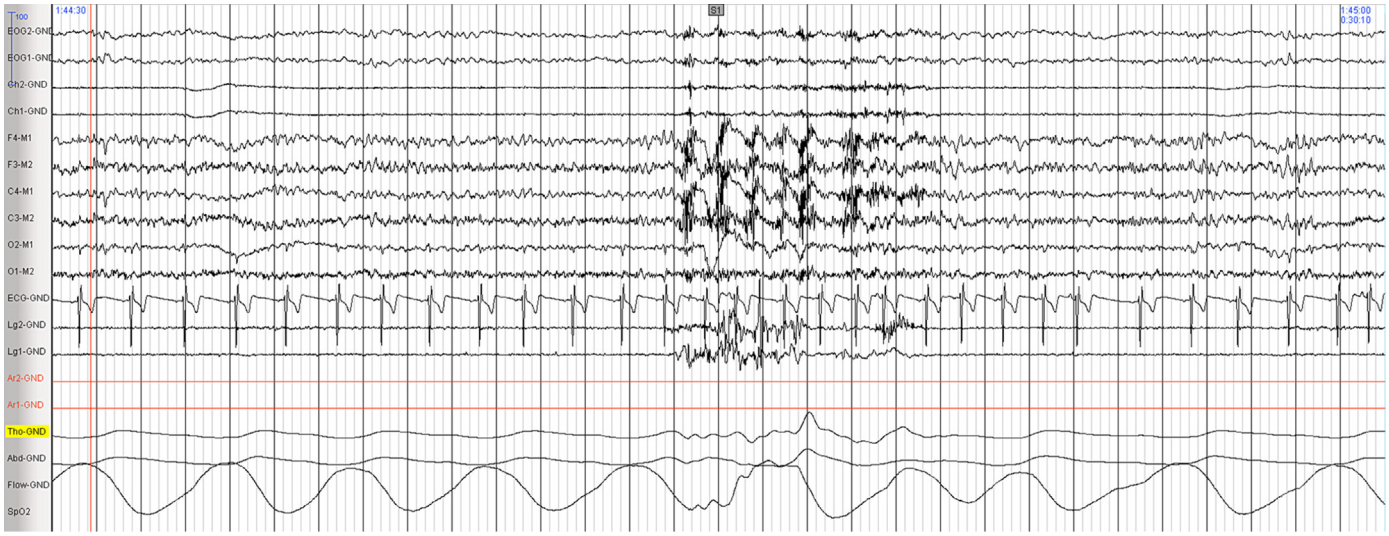

Fig. 1. Benign sleep bruxism episode detected during polysomnography recording (30-s epoch).

The initial treatment with 500 mg levetiracetam administered twice daily was only partially effective, and it was therefore switched to 500 mg valproate acid in two daily doses that was highly effective in suppressing both types of seizures, GTCS and absences. Occlusal splint for sleep bruxism and escitalopram 20 mg per day was added, and our patient underwent psychotherapy focused on stress-management and insomnia. As a result, he currently reports a significant improvement in bruxism, although an examination was not repeated due to his disagreement.

3. Video-analysis

Video 1 shows in detail several seconds of spike-wave complexes, which are immediately followed by a 5-s EMG bruxism pattern accompanied by typical bruxism sound (stage N3, 10-s window, longitudinal bipolar montage). Video 2 shows a tight co-occurrence of spike-wake complexes and 6–7 s sleep bruxism pattern, alternating between each other (stage N3, 30-s window).

4. Brief discussion

In this report, we highlighted the occurrence of sleep bruxism as an epileptic activity-related motor event, which was detected by video-PSG/EEG monitoring. Our patient has several risk factors for benign sleep bruxism (ie, anxiety disorder, chronic stress, insomnia, smoking, drinking alcohol, marijuana use, hereditary influences). Moreover, the examination also revealed more prolonged episodes of bruxism in relation to epileptiform discharges.

To our knowledge, there have been only two cases of tooth grinding reported as epilepsy-related motor events in literature, both in patients with temporal lobe epilepsy [6,7]. The authors concluded that tooth grinding in their cases represented a specific type of epilepsy-related oromandibular automatism rather than a benign sleep bruxism, because it was associated with rhythmic right temporal discharges and disappeared when the seizures were controlled after surgical treatment of epilepsy. Therefore, a direct epileptic mechanism was assumed. Our patient also showed a tight co-occurrence of epileptic discharges and sleep bruxism activity, alternating between each other, but the pathophysiology in our case might be different from the mechanism observed in temporal lobe epilepsy.

We believe that in our case, rhythmic jaw muscle activity is set in motion by both sleep-related mechanisms and epileptic activity. Benign sleep bruxism has been enhanced by sleep instability with highly fluctuating arousal due to the previously mentioned factors (anxiety, alcohol, smoking, marijuana) and concurrently, the epileptic activity could act as an internal trigger increasing arousal instability that in turn could enhance and modulate the occurrence of bruxism by disinhibiting central pattern generators. Conversely, resulting sleep instability could facilitate the occurrence of epileptic discharges in such a bidirectionally influenced system [8–10].

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Conflict of interest

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: https://doi.org/10.1016/j.sleep.2019.06.009.

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