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Can bruxism be a sign of the temporal lobe epilepsy? Case report of a child with bruxism and hippocampal cystic lesion

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Resumo

O bruxismo é uma condição multifatorial e de elevada prevalência na infância. Entretanto, como sinal de epilepsia do lobo temporal, parece ser um evento raro. Há registros de pacientes com bruxismo relacionado a crises do lobo temporal, demonstrando que, excepcionalmente, um evento como o ranger de dentes, pode não ser apenas uma parassonia. Apresentamos uma criança com bruxismo, cuja ressonância cerebral mostra lesão cística, compatível com cisto de fissura coróidea, que comprime o hipocampo esquerdo. O bruxismo foi documentado por polissonografia, com montagem eletrencefalográfica ampliada, sem qualquer expressão eletrográfica durante os episódios. Mostramos assim que a relação de bruxismo e epilepsia neste paciente com lesão potencialmente epileptogênica é remota.

Palavras-chave: Bruxismo; cisto de fissura coróidea; epilepsia do lobo temporal; polissonografia.

Abstract

Sleep bruxism is a condition of high prevalence in childhood and has multifactorial causes. However, as a sign of temporal lobe epilepsy, it seems to be a rare event. There are reports of patients with bruxism related to temporal lobe seizures, showing that, exceptionally, an event such as teeth grinding may not be just a parasomnia. We present a child with bruxism, whose brain resonance shows cystic lesion compatible with choroid fissure cyst, which compresses the left hippocampus. Bruxism was documented by polysomnography, with extended electroencephalographic setup, without any electroencephalographic expression during episodes. Thus, we showed that the relation between bruxism and epilepsy in this patient with potentially epileptogenic lesion is remote.

Keywords: Bruxism; choroidal cyst; temporal lobe epilepsy; polysomnography.

INTRODUCTION

Sleep bruxism is a condition with high prevalence in childhood and has multifactorial causes[1,2]. The majority of sleep bruxers present no associated medical or psychiatric conditions. In these cases, the condition is called primary sleep bruxism. Secondary sleep bruxism has been reported in patients with psychiatric disorders or neurological conditions, including brain injury[2]. However, as a sign of temporal lobe epilepsy, it seems to be a rare event.

The clinical signs of the temporal lobe epilepsy vary considerably: formed hallucinations, illusions, dyscognitive experiences (déjà vu, dreamy states, depersonalization), affective states (fear, depression, or elation), automatism (ictal and postictal). The psychomotor automatisms are the most prominent aspect of complex partial seizures. Automatisms that have been identified include oroalimentay automatisms, such as chewing, lip-smacking, and swallowing; automatism of mimicry, such as fear, anger, anxiety, and joy; gestural automatisms, such as clapping, scratching, and stereotyped repetitive utterance[3].

Teeth grinding was recently included among the manifestations that can describe oroalimentary automatisms

observed during epileptic seizures, caused by abnormal temporal epileptiform discharges, involving limbic structures. Physiopatohologic speculation may include the activation of common central pattern generators involved in the regulation of masticatory rhythms in different physiologic or pathologic conditions[4].

We present the case of a child with bruxism, whose resonance shows cystic lesion compressing the left hippocampus, and discuss the correlation between clinical, electrophysiological and imaging findings regarding the possibility of epilepsy.

CASE REPORT

A male 5-year-old child, born by cesarean section at 32 weeks gestation indicated by fetal centralization. Apgar 7 and 9 in the first and fifth minutes, respectively. Weight: 1200 g (preterm infant, adequate size for the gestational age). The baby was born with right renal agenesis and hypospadias.

He stayed in the NICU for seven days, due to the hyaline membrane disease and neonatal jaundice, was given mechanical ventilation and exchange transfusion, and matured with proper neuropsychomotor development. At the age of nine months, teeth grinding was noticed during sleep. He presented disturbed sleep, but with no automatisms, apnea or periodic limb movement disorder. There is no family history of parasomnia, epilepsy or any other possibly related disorders. Neurological care was sought to assess bruxism and concentration difficulty. He presented night teeth grinding since long ago, which resulted in significant tooth wearing and fracture of incisal edge 21. The patient does not present diurnal bruxism or epileptic seizures and is under dental attendance, using a device interocclusal retention with the purpose of minimizing tooth wearing. He is under multidisciplinary follow-up. Physical neurological exam showed dyslalia. Evolutional neurological evaluation was normal. He underwent neuropsychological assessment, which showed selective attention deficit, with intellectual performance within the average range.

He underwent brain magnetic resonance, which showed a hyper-intense elliptical cystic lesion, measuring $0.7 \times 1.1 \times 1.2$ cm, located in the left choroid fissure, compressing and distally displacing the portion corresponding to the left hippocampus; this finding supposedly represents a choroid fissure cyst (Figures 1-4). The right hippocampus and other cerebral structures are normal.

Extended EEG was performed during sleep and awake, and its result was normal. Patient underwent polysomnography with video-electroencephalographic (Video-EEG) monitoring, according to the international system 10-20. During the polysomnography record the surface electromyography electrode, placed on the face over the bilateral masseter muscles, recorded transitory motor signs lasting up to 20 seconds, confirming the existence of bruxism. The electroencephalography traces during sleep showed normal grapho-elements in the several sleep stages, without any record of an abnormal electrographic activity, including during bruxism.

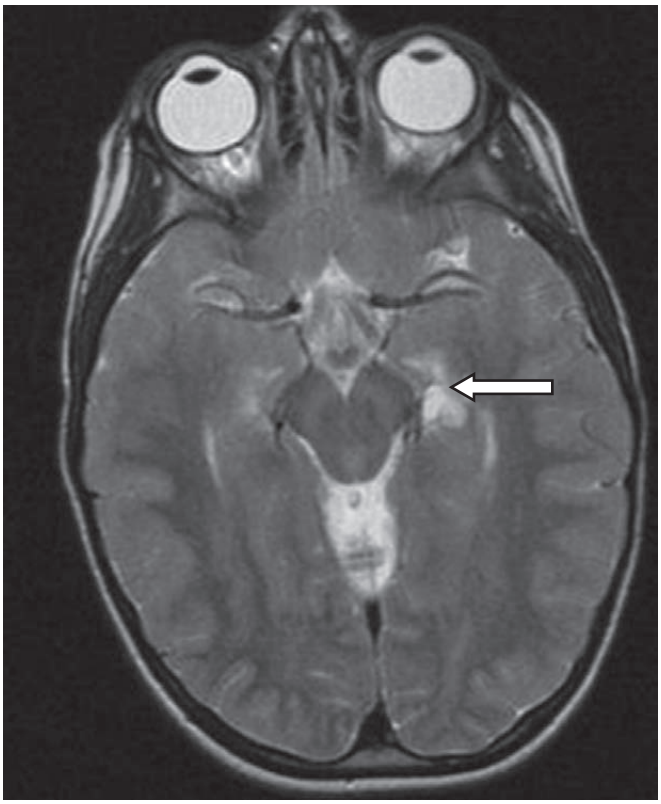

Figure 1. Axial T2. Magnetic resonance demonstrates a cystic (hyperintense) lesion (arrow) in the left choroidal fissure.

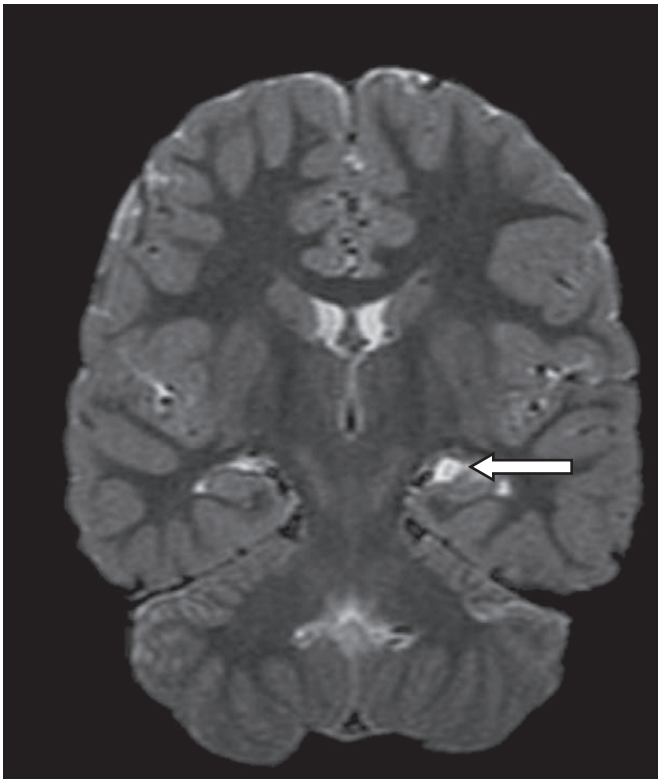

Figure 2. (Coronal T2) Magnetic resonance demonstrates a cystic hyperintense lesion in the left choroidal fissure, located between the fimbria of the hypocampus and the diencephalon.

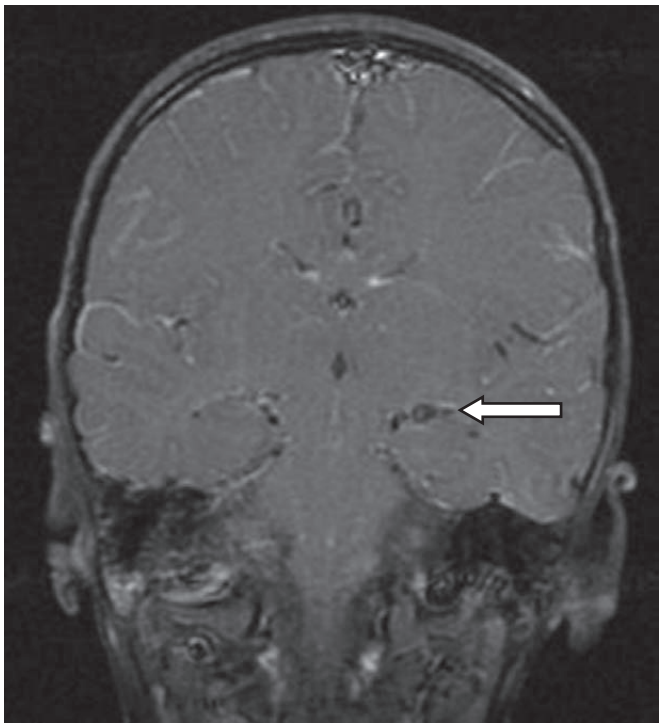

Figure 3. (Coronal T1 after gadolinium). Magnetic resonance reveals a cystic hypointense lesion in the left choroidal fissure, without contrast enhancement.

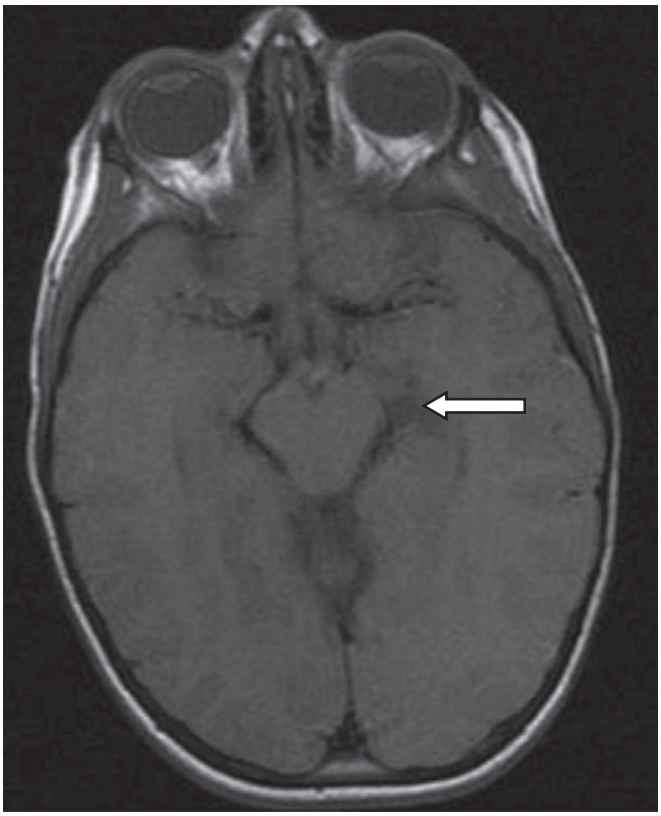

Figure 4. Unenhanced axial T1 shows an ovoid cystic hypointense lesion in the left choroidal fissure, a benign neuroepitelial cyst.

DISCUSSION

Bruxism is a condition, which is not well understood regarding etiology and occurs at any age during stage 2 NREM or REM sleep. The physiopathology of bruxism is unknown, even though stress and anxiety may be considered as risk factors. Genetic and upper airway resistance factors are being investigated[2,5,6].

There are suggestions that generators are triggered during bruxism, involving regulation of the rhythmic chewing movements, whether of a physiological nature or by physiopathologic factors, as in temporal lobe epilepsy. The triggering factor may be an increase in the awakening level, which occurs in the sleep bruxism or occasionally the appearance of epileptiform discharges[4,7,8]. Up to one fifth of the population may be affected. It may be appropriate to "eliminate the possibility of" nocturnal seizures, although, as a sign of temporal lobe epilepsy, it seems to be a rare event[9,10].

We present a child with bruxism, whose MR shows a cystic lesion compatible with choroid fissure cyst, which compresses the left hippocampus, a region that tends to develop epilepsy. Regarding the distinction of parassomia from seizures, it is sufficient to be aware of the diagnostic criteria for each of these disorders and the problems they raise in conjunction with epilepsy.

The main differentiating features characterizing nocturnal seizures are: onset at any age, several attacks per night at any time during the night, brief duration (s) with stereotyped motor pattern. Parasomnias usually start under 10 years, the duration of episodes is variable and may last up to 30 minutes and tend to fade with time. In general, these patients have a positive family history. Further investigations on pathophysiology, genetics and epidemiology are needed to clarify the relationship between epileptic and non-epileptic sleep related paroxysmal phenomena[9,10].

Clinically, temporal lobe epilepsy in childhood manifests itself by a predominance of simple partial seizures; however, with age seizures tend to become complex and less frequent. Motor signs are usually prominent, even though less complex than in adults and with predominance of oral compared to manual automatisms[3,11].

Children younger than four years old presented clear motor signs with tonic and myoclonic components, while, above that age range, patients with exclusively hypomotor seizures would be found[11]. The patient's motor symptoms began at nine months, leading to the suspicion that they were oral automatisms of an epileptic nature, secondary to the hippocampal lesion. Despite the limitations and age range of our patient, it seems to be no doubt that the clinical signs are restricted to the teeth grinding movement. There have not been reports of auras or other gestural, oroalimentary and deambulatory automatisms.

Differently from the etiology in adults (hippocampal sclerosis), temporal lobe epilepsy in children usually is secondary to low-grade tumors and malformation of the cortical development[3]. Morioka et al.[12] described the case of two patients with small choroid fissure cysts, compressing the hippocampus and leading to complex partial seizures.

Even though some series have reported this finding as incidental[13], the choroid fissure cyst seems to be related with focal epileptic seizures. Foa Torres et al.[14] observed that, out of 10 patients with choroid fissure arachnoid cysts, five presented signs of previous focal epilepsy, mainly complex partial seizures.

In a study[4] in which 112 patients were analyzed, monitored by video EEG, bruxism was found as an ictal motor sign in only one patient, evidencing that, exceptionally, an event such as teeth grinding may not be just a parasomnia. This patient presented, besides bruxism, expression of fear and hypomobility during the seizure and the VEEG record showed left anterior temporal epileptiform activity, with reversal phase in electrode F7, followed by brief EEG attenuation. The MR showed hippocampal atrophy with sclerosis, confirmed by the pathology. In such a case, the teeth grinding act represented oroalimentary automatism, occasionally induced by epileptiform discharges involving limbic structures. This observation was a warning as to the possibility that the bruxism presented by our patient could correspond to a sign of temporal lobe epilepsy.

The extended interictal EEGs, taken awake and asleep were normal, which, however, does not discard the possibility of epilepsy. It is a fact that video EEG recording is the golden standard for topographic definition of the epileptogenic focus[15]. Therefore, along with the polysomnography, video-EEG monitoring was performed. Even during recording of motor activity secondary to bruxism, the electrographic tracing showed only muscular artifacts, which makes it unlikely to correlate the symptom presented with the occurrence of epilepsy, although deep electrodes were not been placed.

Hence, despite the existence of a potentially epileptogenic lesion compressing the hippocampus, we were not able to establish a relation between bruxism and temporal lobe epilepsy in our patient. Essential historic elements that distinguish these events and the role of video-EEG-polysomnography in their differential diagnosis are emphasized.

CONCLUSION

Bruxism is a frequent sleep disorder mostly not epileptic at all; a good clinical history is generally sufficient to make the right diagnosis. In the case reported in this paper, the association with a focal lesion may justify a sleep EEG recording and also a video EEG recording, that is dispensable in the majority of sleep bruxers.

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