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Short communication

Severe bruxism following basal ganglia infarcts: insights into pathophysiology

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Abstract

Bruxism characterized by clenching and grinding of teeth can lead to toothwear, headaches and depression. While bruxism has been associated with a number of neurological diseases, it has not been highlighted following cerebral infarction.

An elderly man presented with an acute onset of tooth grinding and jaw clenching associated with dysarthria. His bruxism was worse during the day and resolved during sleep. He had frequent jaw aches, headaches and swallowing difficulty. Examination demonstrated the presence of dysarthria with jaw clenching and tooth grinding, producing persistent high pitch and loud squeaky sounds. A magnetic resonance imaging and angiography examination revealed a recent infarct in the right thalamus. In addition, chronic lacunar infarcts were present in the bilateral caudate nuclei with severe basilar artery stenosis. He was successfully treated with botulinum toxin.

We discuss the pathophysiologic mechanisms of bruxism associated with basal ganglia infarcts. Dysfunction of the efferent and/or afferent thalamic or striatopallidal tracts may play a role in bruxism. Early recognition of bruxism following stroke could reduce unnecessary suffering since the condition can be effectively treated.

Keywords: Thalamic infarct; Bruxism; Botulinum toxin

1. Introduction

Bruxism refers to the presence of diurnal or nocturnal parafunctional activity such as clenching, grinding, bracing and gnashing of the teeth [1]. Its reported prevalence in the adult population is highly variable, presumably from differences in diagnostic criteria [1–5]. Severe bruxism can cause tooth damage, temporomandibular disorders, swallowing and speech difficulties, headaches and depression. Such problems can complicate stroke rehabilitation. Bruxism has been observed in brain injuries, neurodegenerative diseases and mental retardation, though the exact etiology of nocturnal bruxism, the most common type in the general population, remains unknown [1–5].

Pathophysiologic studies suggest underlying dopaminergic system dysfunction [2–4]. We highlight the first report of a patient who developed severe bruxism following a recent thalamic and chronic caudate infarcts, and discuss the involvement of the basal ganglia in the pathogenesis and the role of botulinum toxin in modifying the course of bruxism. We also draw attention to the observation that bruxism following basal ganglia strokes maybe an unrecognized complication.

2. Case report

The patient, a 64-year-old man with a long history of hypertension and hyperlipidemia, had presented with non-vertiginous dizziness and left-sided weakness. Neurological examination revealed mild dysarthria and left hemiparesis of grade 4+/5. A brain magnetic resonance imaging and angiography (MRI/A) examination showed lacunar infarcts in the head of both caudate nuclei, both external capsules

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and left lentiform nucleus, with severe basilar artery stenosis (Fig. 1, upper row). He was anticoagulated with warfarin with gradual resolution of his neurologic symptoms.

About one and a half years later, he presented with an acute onset of tooth grinding and jaw clenching associated with dysarthria. Initially, he could voluntarily suppress these jaw movements for a few seconds, but subsequently these movements progressed in severity to the extent that they became constant and persistent during the day. The loud grinding sounds created much distress to him and his family members. He also complained of frequent jaw aches and headaches. His bruxing episodes disturbed his swallowing and he was socially embarrassed and depressed. However, the bruxism usually resolved during sleep and he had no early morning jaw pain. There was no previous or family history of bruxism, no history of psychiatric illness or exposure to neuroleptics, antidepressants, sedative or anxiolytic medications or drugs of abuse.

Neurologic examination demonstrated the presence of jaw clenching and tooth grinding, producing persistent high pitch and loud squeaky sounds. There was no significant masseter hypertrophy but some degree of tooth wear was present. There were no involuntary orolingual movements or evidence of movement disorders elsewhere. He had no focal neurological deficits, except for dysarthria. A repeat MRI/A revealed an interval new lacunar infarct in the right thalamus (Fig. 1, lower row).

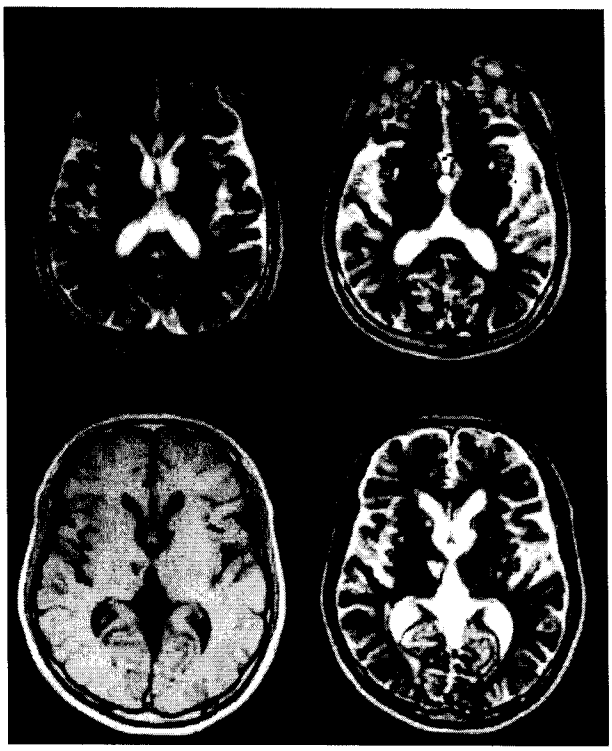

Fig. 1. Consecutive axial T2-weighted images demonstrating lacunes in both external capsules and caudate head nuclei (upper row). MRI 1.5 years later shows interval new right anteromedial thalamic lacunar infarct on T1-weighted and T2-weighted images (lower row).

He was initially treated with clonazepam with little improvement of his bruxism. The patient was subsequently treated with *Dysport*® (Speywood Biopharm, UK), where 25 units were injected at two to three sites intramuscularly into each side of the masseter muscles. Within a month, the patient reported some improvement of his bruxism. A second injection with *Dysport*® (50 units per side) 3 months later provided greater relief to his symptoms. Subsequently, he required lesser botulinum toxin (BTX) injections as the severity of his bruxism without treatment was much improved.

3. Discussion

To our knowledge, this is the first report of severe bruxism associated with cerebral infarction. Our patient's bruxism was most likely precipitated by the acute thalamic infarct and/or contributed by the chronic caudate infarcts for the following reasons: (1) his bruxism onset was acute; (2) there were no previous or family history of bruxism and movement disorders; (3) he had no other neurological and systemic illness, or drug exposure to possibly account for his symptoms; (4) MRI showed a new thalamic infarct compared with baseline scans; and (5) the diurnal pattern of his bruxism with no nocturnal symptoms was compatible with the clinical description of secondary bruxism, different from the common variety of bruxism, which happened during sleep [1–5].

The pathophysiology of bruxism is still debated. Older theories such as occlusal discrepancies with reflex contraction of the jaw muscles have frequently been challenged [1,2]. It is possible that there exists a central "bruxism generator" complex, which helps modulate the interactions of the motor, limbic and autonomic systems, and control the pattern of bruxism [2]. The theory of dopaminergic dysfunction in bruxism patients can be supported by some of the following observations: (1) reports of bruxism in patients with Huntington's disease [6] and cranio-cervical dystonia [4]. A higher than expected frequency of bruxism and jaw movements have also been reported in Parkinson's disease compared to controls [4]. (2) Clinical efficacy of dopaminergic drugs such as bromocriptine and levodopa in alleviating bruxism [7–9]. (3) Induction of non-functional masticatory activity in rats by repeated stimulation of the dopaminergic system with apomorphine [10]. (4) Demonstration of an abnormal side imbalance in striatal D2 receptor expression on functional imaging in patients with nocturnal bruxism [11].

The thalamus, which receives afferents from the striatum, is the main gateway of the motor outflow tracts from the basal ganglia. Sensory afferents from the trigeminothalamic tract also terminate in the thalamus. The right thalamic

infarct in our patient was predominantly within the territory of the paramedian thalamic–subthalamic arteries. A review of the literature shows that infarcts within this area are associated with a change of consciousness, neuropsychological and vertical gaze disturbances [12]. None of these signs were found in our case. Delayed onset movement disorders such as tremor and dystonia have been reported with thalamic and other static brain lesions [12–14]. A syndrome of complex movement disorders have also been described in infarcts of the postero-lateral and paramedian thalamus, presumably by disruption of the motor outflow tracts to the motor cortex [15].

How could thalamic or caudate infarcts possibly cause bruxism? In animal studies, stimulation of the limbic structures or the jaw area of the motor cortex can produce bruxism [16]. In our patient, it is likely that dysfunction of the afferent and/or efferent pathways to the thalamus as a result of the infarct lead to a disruption of the normal interplay of neuronal circuits connecting the striatum, thalamus, subthalamus and cerebral cortex. This in turn created a loss of inhibition of the higher cortical control on the trigeminal motor nuclei located in the pons. The thalamic infarct could also be a triggering event for the bruxism in a background of dopaminergic dysfunction as a result of the chronic caudate infarcts. Transcranial magnetic stimulation studies in human subjects during different biting tasks have demonstrated that the corticotrigeminal projections to masseter are bilateral, with a stronger contralateral projection [17]. This can explain why a right thalamic infarct in the patient could lead to bruxism involving both masseters. In support of this, animal studies have also showed that inputs from the premotor area to the trigeminal nuclei and their connections within the pontine reticular formation are bilateral but with ipsilateral preponderance. The neuronal circuits within the pontine reticular formation and the trigeminal and facial nuclei play an integral part in oral motor behavior [18,19]. The caudate infarcts could result in variable disruption of both the direct and indirect striato-pallidal pathways leading to an altered thalamocortical drive.

It remains speculative why thalamic or striatal infarct-induced bruxism appears rare, though a genetic predisposition cannot be discounted. It is possible that the cause and effect of bruxism in stroke patients may be unrecognised especially if the onset of bruxism happened weeks or months after a stroke, or the cause be attributed to dental malocclusion or neuroleptic drugs. Alternatively, it could also be that bruxism following thalamic or basal ganglia strokes are relatively mild in most patients, and hence the awareness of this problem is low.

The effectiveness of BTX in severe bruxism has been highlighted previously [5,20]. However, interestingly, we observed that the toxin lessened the baseline severity of bruxism after only one to two courses of treatment in our patient. It is conceivable that BTX could alter the proprioceptive input to the thalamic nuclei, and hence disrupt the higher cortical efferent input to the trigeminal motor nucleus.

In conclusion, we highlighted an unusual case of severe bruxism associated with recent thalamic and chronic caudate infarcts. Dysfunction of the thalamic or striatopallidal tracts may play a role in the pathogenesis of this condition. Early recognition of bruxism can reduce unnecessary suffering as it can be effectively treated. A prospective case control study would be useful to address the prevalence of bruxism following strokes.

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