Delayed-Onset Continuous Bruxism with Olivary Hypertrophy After Top of the Basilar Syndrome

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Delayed-onset continuous bruxism due to brain stem infarction has not yet been reported. A 49-year old man presented with quadriplegia and ophthalmoplegia. Brain MRI showed acute infarction in the bilateral midbrain, right thalamus and the superior cerebellum. One month later, the patient developed bruxism which persisted during sleep. A palatal myoclonus was not observed. Follow up MRI taken 4 months later showed bilateral olivary hypertrophy. We suggest that the patient's bruxism may be related to the olivary hypertrophy. The bruxism generator may be located in the pontine-reticular-formation (PRF). Bilateral large midbrain lesions interrupting the cortical inhibition may have produced bilateral olivary hypertrophy, which could stimulate the PRF, producing continuous bruxism.
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Bruxism is characterized by parafunctional stereotyped rhythmic activity of the masseter muscles. Sleep bruxism is a common condition, being present in 90% of the population in their life time.[1] Symptomatic daytime bruxism has been reported in patients who had basal ganglia infarction, multisystem atrophy or used amphetamine.[2-4] Olivary hypertrophy is related to delayed continuous palatal myoclonus and develops a few weeks after the dentato-rubro-olivary pathway is damaged. Brain stem lesions with palatal myoclonus and olivary hypertrophy are common. However, to the best of our knowledge, bruxism with olivary hypertrophy due to a brain stem lesion has not been reported to date. In this report, we describe a patient showing delayed-onset, continuous bruxism with olivary hypertrophy after top of the basilar syndrome and discuss possible pathophysiolgic mechanisms.

CASE REPORT

A 49-year-old man with a history of sick sinus syndrome presented with quadriplegia and ophthalmoplegia. Brain magnetic resonance imaging (MRI) and an angiography examination, which were performed a day after quadriplegia, revealed acute brain infarction in the bilateral midbrain, right median thalamus, and superior cerebellum associated with the distal basilar artery occlusion. One month later, he developed bruxism (80 cycles/min). This symptom persisted during daytime and during sleep. He was treated with L-dopa and baclofen, which were not effective. Palatal myoclonus

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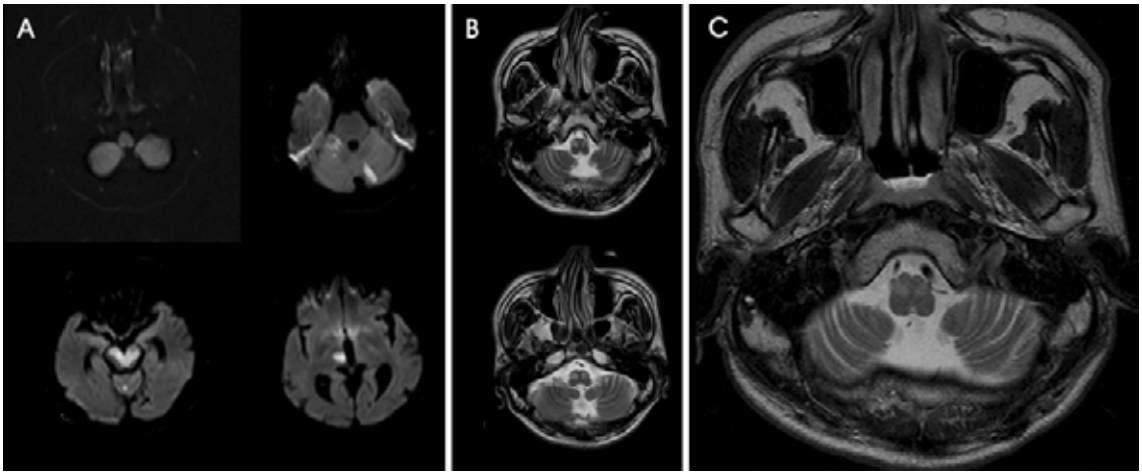

Figure 1. MRI findings at the onset of infarction (A). Acute infarction was observed in the bilateral midbrain, right median thalamus, and superior cerebellum. Follow-up T2-weighted MRI performed four months later revealed bilateral olivary hypertrophy (B). T2-weighted MRI performed eight months after the infarction revealed persistent olivary hypertrophy (C).

was not observed. Four months after quadriplegia, he continued to experience bruxism. Follow-up MRI revealed bilateral olivary hypertrophy on a T2-weighted image. An MRI examination performed eight months later revealed persistent olivary hypertrophy. The bruxism ceased two years after the stroke onset (Fig. 1).

DISCUSSION

The mechanism of bruxism remains unclear. It is also unclear whether the mechanism of symptomatic bruxism is identical with that of idiopathic sleep bruxism. It has been shown that rhythmic jaw movement is generated by a neuronal activation in the brainstem of animals.[7] Rhythmic firing of interneurons in the pontine reticular formation (PRF) and a connection between the oral motor nuclei and the PRF have been observed.[7-10] Rodents move their jaws continuously and cows ruminate throughout the day. However, there is no rhythmic jaw movement in humans under normal conditions. We suggest that the bruxism generator is located in the PRF. The bruxism center in the PRF is normally suppressed by high cortical inhibition. However, the activity of the generator is so weak that the release from high cortical inhibition during normal sleep may not be sufficient to cause bruxism. Thus, the release phenomenon due to sleep and other factors such as dental problems, smoking, alcohol, and the coma state[6] may cause bruxism.

In our case, bilateral midbrain infarction may have caused the interruption of higher cortical inhibition and damaged the dentato-rubro-olivary pathway. Although higher cortical inhibition is interrupted, the brainstem generator may not be sufficient to cause bruxism. Additional damage to the dentato-rubro-olivary pathway may cause bilateral olivary hypertrophy, which can stimulate the PRF. Because no connection between the oral motor nuclei and the inferior olivary nuclei has been observed in animal study, we do not think that the hypertrophied olivary nucleus directly stimulates the motor nucleus in the brainstem.[8] Instead, hyperactive bilateral PRFs may cause bruxism. Pollack and Cwik have reported bruxism after cerebellar hemorrhage,[5] which may also be related to the damage to the dentato-rubro-olivary pathway.

Unlike palatal myoclonus, bruxism requires bilateral harmonic rhythmic contraction and relaxation of involved muscles. Since the neurons in the PRF project to both the trigeminal motor nuclei,[8] the PRF may induce the contraction of one side and relaxation of the opposite side. On the other hand, the normal side can influence both the trigeminal motor neurons, and bruxism may require a bilateral PRF change. We suspect that this is a reason why bruxism is very rare in patients with

brainstem infraction.

In conclusion, we report an unusual case of continuous bruxism with olivary hypertrophy after brainstem infarction. Furthermore, we suggest that the human bruxism generator may be located in the PRF. Large bilateral midbrain lesions that interrupt cortical inhibition seem to produce bilateral olivary hypertrophy, resulting in bruxism in the presence of the intact pontine generator.

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