Jaw Dystonia Induced by Speaking

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We describe a 43-year-old housewife who presented with dysarthria suddenly because her masseter muscles contracted bilaterally, when she was speaking. Brain MRI showed focal signal change on midbrain. Jaw dystonia induced by speaking is very rare and we chose an anticholinergic medication, rather than botulinum toxin injection. Her condition was markedly improved after medication. We suspected that her symptoms were related with focal lesion, so she had secondary jaw dystonia induced by speaking. 

Key Words: Jaw dystonia, Speech, Infarction

Jaw dystonia, a form of oromandibular dystonia, is involuntary contraction of the masticatory muscles, resulting in dysarthria or dysphagia. Jaw dystonia induced by speaking is very rare and has not been described in association with structural lesions. We recently examined a patient who developed sudden onset jaw dystonia induced by speaking, probably caused by a lesion in the midbrain.

CASE REPORT

A 43-year-old, right-handed housewife experienced a sudden-onset speech disturbance. She was a devout believer and she used to pray in a loud voice. Two days before admission to our hospital, while shouting her prayers, she suddenly could not speak properly because of difficulty in opening her mouth. She was conscious of contracture of her masseter muscles bilaterally, but felt no pain in the facial area, including the temporomandibular joints. When we first examined her, she had no other symptoms of dystonia. She had no history of medication or facial injury. Her family history was not remarkable for neurological disease. She complained only of difficulty opening her mouth while speaking, but she felt comfortable and no problem to speak when she was eating or chewing gum. Consequently, she would take gum or candy with her when she went visiting. Her physical and neurological examinations revealed speech disturbance only. When she was talking, upper and lower teeth stuck to each other, but not lips. Her maseter muscles excessively contracted bilaterally. Her teeth were normal and there were no abnormalities of the temporomandibular joint area, which was examined in the dental department. Laboratory findings, including thyroid function tests, serum copper and ceruloplasmin, and CSF examination, were within normal limits. Electroencephalography was normal, but brain magnetic resonance imaging showed a high signal intensity in the left substantia nigra area of the midbrain on T2 weighted imaging (Fig.). When we first examined her, we suspected a psychogenic movement disorder due to the sudden, bizarre symptoms, so a psychologist interviewed her, and conducted depression and personality tests, but there were no abnormal findings. She was given a placebo injection, but there was
no difference in her symptoms before and after injection. She was treated with anticholinergic medication, trihexyphenidyl 4 mg/day, and antiplatelet medication, clopidogrel 75 mg/day. When she was taken initial medication, her mouth started to open gradually on speaking. Five months after initiating the medication, the dosage of trihexyphenidyl was gradually tapered because her symptom was improved. There was no speech impairment from dystonia without medication one year later.

**DISCUSSION**

Oromandibular dystonia, a form of focal dystonia, affects the masticatory or tongue muscles, causing dysarthria or dysphagia due to involuntary jaw or tongue movements. Jaw dystonia is a rare disorder, in which the tone of the masseter or temporalis muscles is increased during resting and actions including eating, chewing, and speaking. This focal dystonia may result from neuroleptic drugs, orofacial injury, or unknown causes. Our patient had no past history of orofacial injury, neuroleptic medication, or dental problems. There are two characteristics on this case. One is that her symptoms occurred under specific situation, and the other is that she experienced her symptoms suddenly and brain MRI showed suspicious lesion. In a previous study, patients could not open or close their mouth because their symptoms continued whether or not the jaw was at rest. These patients had hypertonus of the target muscles, including the masseter or temporalis muscles, and improved dramatically with the injection of botulinum toxin in the target muscles. We did not perform an electrophysiological study because our patient was normal at rest. After considering the side effects of botulinum toxin, we chose trihexyphenidyl, because she had a minor speech disturbance induced only by speaking. Some authors reported similar cases before, but their cases were idiopathic and had lingual dystonia, not jaw dystonia.

Rarely, focal facial dystonias, including blepharospasm or jaw closing or opening dystonia, have been described in association with structural lesions. These structural lesions include those of the bilateral basal ganglia, rostral midbrain in blepharospasm, parietal lobe in blepharospasm and jaw closing dystonia, and pontine lesion in jaw opening dystonia. Our patient’s symptom occurred suddenly and, unfortunately, diffusion weighted brain MR imaging was not available at that time. Brain MRI showed a lesion, probably an acute infarction, in the substantia nigra near the red nucleus area, as described above (Fig.). Despite suspicious evidence supporting the relationship between midbrain infarction and subsequent development of jaw dystonia, especially that induced by speaking, the pathophysiological mechanisms are not well understood.

Many patients improved with injection of botulinum toxin, as reported for other focal dystonias. In minor focal dystonia, especially that induced by speaking, as in our patient, we suggest that trihexyphenidyl is a good initial treatment, as suggested by others.
LEGENDS TO THE VIDEOTAPES

Segment 1. On examination, the patient has difficulty opening her mouth during speaking.

Segment 2. Eight months after initiating medication, the patient is free of jaw dystonia and her mouth opens during speaking.

REFERENCES
