

When Drinking Makes the Tremor Worse: A Task-Specific Orolingual Tremor

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The patient was a 49-year-old right-handed female homemaker. She reported a 6-month history of movements affecting the mouth and jaw when drinking. It was not present when eating. She had noticed that the movements were not precipitated by drinking with a straw, and she had taken to drinking in this fashion. Stress and caffeine were not exacerbating factors. The movements seemed to be reduced by drinking at least three glasses of wine, but she considered this intake excessive. There was no history of tremor or abnormal movement in the hands, arms, head, voice, or legs. There was no stiffness or slowness of the limbs nor walking difficulty. There was no reported disturbance of sleep and no history of headache or jaw pain suggestive of bruxism. She had played trumpet for 8 years while at high school. There was no history of oral trauma or recent dental procedure.

There was no relevant past medical history. Her maternal grandmother had a tremor when holding items in her right hand, but no tremor at rest, and there was no family history of Parkinson’s disease or essential tremor.

On examination, there was no visible tremor of the jaw, lips, head, or limbs at rest. A fine tremor of the jaw and lips became visible upon drinking from a cup (see Video). She could not voluntarily suppress the movement, but tended to take short sips of liquid, reducing the duration of the tremor. Cranial nerves, motor, sensory, cerebellar, and gait examination were also unremarkable. Reflexes were normal.

Complete blood count, hepatic profile, and renal and thyroid function were unremarkable. Inflammatory markers were not increased. Serum manganese, urinary copper, and mercury were within normal limits. MRI brain with contrast showed a normal brain with minimal leukoaraiosis. There was no evidence of restricted diffusion or contrast enhancement.

The patient was only mildly inconvenienced by the tremor and declined further treatment.

Neurophysiological Findings

A multichannel surface electromyographic (EMG) recording was performed using silver/silver chloride electrodes placed 2 to 4 cm apart (Motion Lab Systems, Inc., Baton Rouge, LA). EMG signals were digitized at a sampling rate of 2,000 Hz through a filtered bandwidth of 1 to 1,000 Hz and stored for later analysis. Recorded muscles were bilateral upper and lower orbicularis oris, digastric, masseter muscles, and unilateral forearm flexors and first dorsal interosseus. Temporalis muscle was not recorded, but no activity could be palpated in this muscle during recording. There was no activity in the muscles at rest. Figure 1 shows the pattern of muscle activity upon drinking water from a cup. There is a very regular, 7.5- to 8-Hz tremor, best observed in the upper orbicularis oris muscles bilaterally. It was not present in the other muscles studied. The tremor immediately disappeared when drinking ceased. No abnormal muscle activity could be observed or recorded with any position of the mouth, lips, or jaw, including when the patient pursed her lips, mimed drinking from an empty cup, drank with a straw, held liquid in the mouth, held her mouth open, talked, or laughed.

Discussion

We present a patient with an unusual task-specific tremor of the orolingual muscles caused by drinking. There was a family history possibly consistent with essential tremor. Though essential tremor may present with an isolated tremor, task-specific tremor is not a feature.¹ There was no clinical evidence of tremor or dystonia affecting the remainder of the body. Neither coactivation nor reciprocal inhibition can be demonstrated for the orbicularis oris, but the regularity of the tremor observed and the lack of associated neurologic abnormality suggest a task-specific orolingual tremor. The classification scheme proposed

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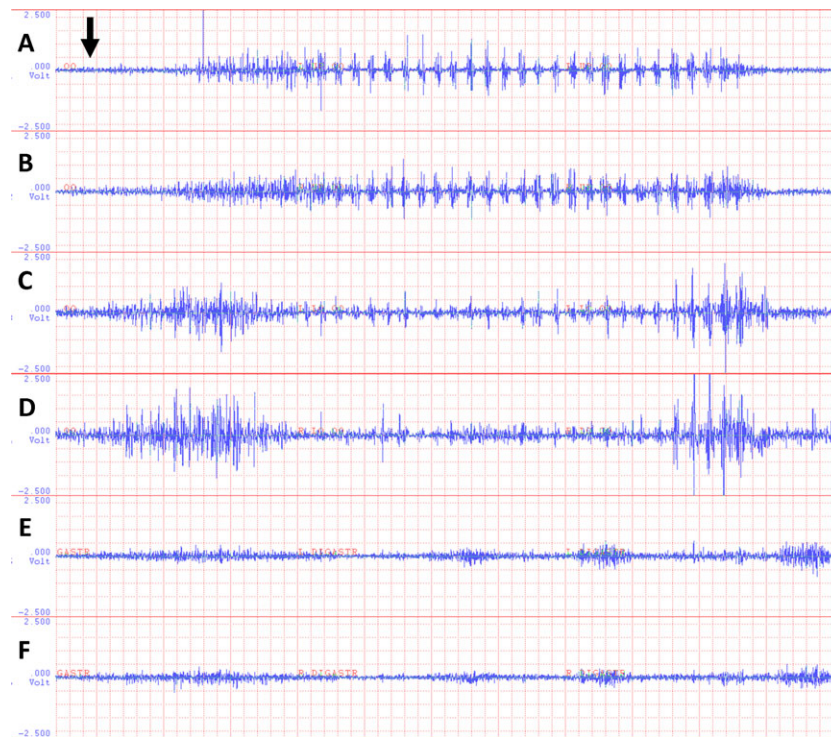


Figure 1 Surface EMG recording. (A and B) Upper orbicularis oris (left, right), (C and D) lower orbicularis oris (left, right), and (E and F) digastric muscles (left, right). Recording shows patient taking a long sip of water and swallowing. Initial pursing of lips with activation of orbicularis oris (black arrow) is followed by onset of a regular 7.5- to 8-Hz tremor affecting orbicularis oris (most marked in the upper pair of electrodes). With cessation of drinking, the tremor stopped. Two contractions of digastric muscle were visible toward the end of the trace, as the patient swallowed.

by Silverdale et al.² requires a task- or position-specific orolingual tremor to occur exclusively during a task or with particular position, without evidence for associated neurological abnormality, particularly dystonia, and in the absence of other known causes of orolingual tremor.

Tremor is defined as rhythmical, oscillatory, and involuntary movements of a body region.³ Task-specific abnormal movements of the orolingual and jaw musculature are rare, but embouchure dystonia (ED) is observed in professional musicians. ED variably affects the muscles of the mouth and jaw required to form the particular position (embouchure) required for playing a woodwind or brass instrument. Onset is generally in the fourth decade of life and the condition can be progressive, spreading to other body regions or other oral functions, such as speaking and drinking.⁴ In a case series of 89 musicians with ED, the most common variant observed was a fast regular tremor of the lips, also termed embouchure tremor (ET).^{4,5} No case of ET developed spread of symptoms to other oral activities. The remote history of trumpet playing in our case is interesting, but of uncertain significance because ET develops in musicians with a long playing history.

Two cases of jaw tremor precipitated by drinking have been described. In each case, the tremor was position sensitive and could be precipitated by jaw position alone.^{6,7} Unique examples of task-specific orolingual tremor include lip tremor precipitated by smiling⁸ or speaking.^{4,9}

Botulinum toxin can be an effective treatment for task-specific orolingual tremor, but is less effective in ED.^{4,5,7}

In conclusion, this patient demonstrated an unusual focal tremor of the lips triggered by drinking that can be categorized as a task-specific orolingual tremor.

Author Roles

(1) Research Project: A. Conception, B. Organization, C. Execution; (2) Statistical Analysis: A. Design, B. Execution, C. Review and Critique; (3) Manuscript Preparation: A. Writing of the First Draft, B. Review and Critique.

C.O.G.: 1A, 1C, 3A

J.H.B.: 1A, 3B

J.M.: 1A, 3B

O.K.: 1A, 3B

N.K.: 1A, 3B

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Supporting Information

A video accompanying this article is available in the supporting information here.

Video. Segment 1: patient shown at rest, speaking (00:00–00:20). Segment 2: patient shown drinking (00:21–01:32). Segment 3: patient shown miming drinking (01:33).