

A Case of Intractable Psychogenic Essential Palatal Tremor

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Essential palatal tremor (EPT) is a rare disorder which shows rhythmic involuntary movement of the muscles of soft palate, especially tensor veli palatini muscle. EPT is classified by two subtypes, which is primary and secondary EPT. Secondary EPT includes psychogenic type. We describe a case of intractable psychogenic EPT.

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Palatal tremor (PT) or palatal myoclonus is a movement disorder characterized by rhythmic movements of the soft palate at 0.5 to 3 Hz.¹ PT is classified as two types of essential (EPT) and symptomatic (SPT). EPT generally affects children of both genders, whereas SPT is most commonly observed in adult males.² EPT is bilateral and usually disappears during sleep, whereas SPT is more frequently unilateral and persists even during sleep.² Patients with EPT usually have an ear click, which is absent in the SPT.² The tensor veli palatini (TVP) innervated by the trigeminal nerve is mostly involved in EPT, whereas in SPT the levator veli palatini innervated mainly by the vagus nerve is affected.³⁻⁷ Generally, SPT is most often and due to brainstem or cerebellar disease, but EPT is rare and have no documented brain lesions.¹

Classification of EPT was newly proposed as isolated, indicating the lack of any further signs, and includes primary isolated PT (classical EPT) and secondary isolated PT (PT as a special skill, palatal tic, and psychogenic PT).⁸ In the recent study, the etiology of EPT was found to have psychogenicity in 70% of the patients with published criteria for diagnosis of psychogenic movement disorders (PMDs).⁹ Therefore, psychogenic PT may be underrecognized and not uncommon.⁹ Herein, we report a case of a patient with intractable psychogenic EPT.

Case

A 16-year-old boy visited to our neurology clinic with a 12-month history of a clicking noise in the both ears and palatal discomfort. These symptoms were developed after voice abuse in Karaoke. The clicking resembled the sound of snapping fingernails and was externally audible at a distance of 50 cm. He denied any voluntary control over this movement as well as any sensation of urge before the movement. He complained of throat fullness in daytime.

On neurologic and otolaryngologic examination, rhythmic contraction of whole soft palate muscles was noted. The movement was able to be suppressed for a few seconds by touching the back of his neck, but it could soon return after the voluntary suppression. In addition, the entrainment at each slow and fast finger tapping with external pace was found (Video).

The remaining physical examination, development, and past history were unremarkable.

Needle electromyography (EMG) of TVP muscle showed rhythmic contractions with a frequency of 2.6 Hz with amplitude of 150 μ V (Figure 1). There were no abnormal results in other laboratory, audiologic tests, and brain magnetic resonance imaging.

After diagnosis of EPT, despite of medications including benzodiazepines, muscle relax-

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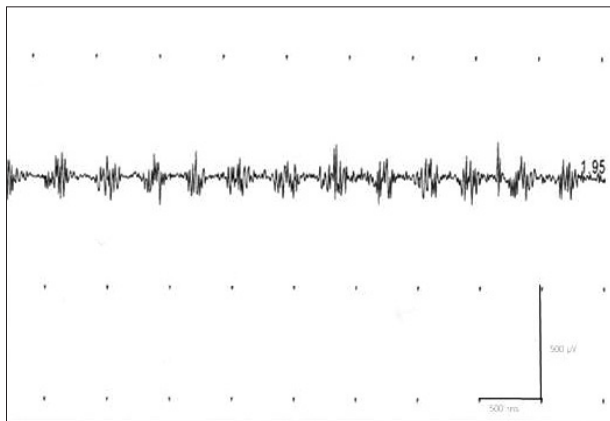


Figure 1. The electromyogram (EMG) of soft palate. The EMG recorded before botox® injection showed rhythmic contraction of tensor veli palatini muscle at firing rate of 2.6 Hz with amplitude of 150 μ V.

ants, and anticonvulsants for 1 month, his symptoms were not changed at all. Therefore, we started to inject 15 units of BTA (Botox®, Allergan, Inc., Irvine, CA, USA) into bilateral soft palates under EMG guidance. Though the success of the first BTA therapy was lasted about 10 weeks, the additional injections of BTA were needed for 3 times due to recurrence. The dosage of BTA was finally elevated to 20 units and there were 4 times injection of BTA at interval of 3 months, but the presenting symptoms showed again.

Discussion

His involuntary movements were clinically closed to EPT because of ear click perceived as objective tinnitus, bilateral involvement, no brain lesions and involvement of TVP muscle by EMG.

Although no single clinical finding is pathognomonic for PMDs, several features are quite helpful.¹⁰ In general, PMDs are characterized by distractibility, entrainment, coactivation sign, variable frequency, amplitude and direction, increase with attention, and poor response to medications.^{8,11} This patient also showed entrainment of soft palate muscle contraction to external paced finger tapping.

Furthermore, PMDs have particular histories including precipitating factor or trivial trauma preceding acute onset, emo-

tional trigger, psychological stressors and psychiatric comorbidities such as anxiety disorders.⁸ In this patient, mild trauma (e.g., voice abuse) before onset suggest to psychogenic type of EPT rather than primary type.

In our case, the only difference from usual psychogenic PT is that this abnormal movement was not well responded to treatment. Psychogenic PT is usually reported to have good response to non-physiological treatment or placebo.⁸

In summary, bilateral objective tinnitus, bilateral TVP muscles involvement, mild trauma before onset, and entrainment of soft palate contraction suggest a psychogenic EPT.

Legend to the Video

Palatal movement of this patient shows entrainment by finger tapping with external pace.

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