A Case of Intractable Psychogenic Essential Palatal Tremor

Eun Joo Chung
Hyun Jung
Sang Jin Kim

Departments of Neurology and Otorhinolaryngology-Head & Neck Surgery, Busan Paik Hospital, Inje University College of Medicine, Busan, Korea

Received September 8, 2012
Revised September 26, 2012
Accepted September 26, 2012

Corresponding author
Sang Jin Kim, MD
Department of Neurology, Busan Paik Hospital, Inje University College of Medicine, 75 Bokji-ro, Busanjin-gu, Busan 614-735, Korea
Tel +82-51-890-6425
Fax +82-51-895-6367
E-mail jsk502@hotmail.com

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.

Key Words: Tremor, Palate, Psychogenic.

Palatal tremor (PT) or palatal myoclonus is a movement disorder characterized by rhythmic movements of the soft palate at 0.5 to 3 Hz. 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. EPT is bilateral and usually disappears during sleep, whereas SPT is more frequently unilateral and persists even during sleep. Patients with EPT usually have an ear click, which is absent in the SPT. 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. Generally, SPT is most often and due to brainstem or cerebellar disease, but EPT is rare and have no documented brain lesions. 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). In the recent study, the etiology of EPT was found to have psychogeneity in 70% of the patients with published criteria for diagnosis of psychogenic movement disorders (PMDs). Therefore, psychogenic PT may be underrecognized and not uncommon. 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 μV (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-
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. In general, PMDs are characterized by distractibility, entrainment, coactivation sign, variable frequency, amplitude and direction, increase with attention, and poor response to medications. 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, emotional trigger, psychological stressors and psychiatric comorbidities such as anxiety disorders. 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.

In summary, bilateral objective tinnitus, bilateral TVP muscle 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.

**REFERENCES**

1. Deuschl G, Mischke G, Schenck E, Schulte-Mönting J, Lücking CH. Symptomatic and essential rhythmic palatal myoclonus. Brain 1990; 113(Pt 6):1645-1672.
2. Margari F, Giannella G, Leccia PA, Fanizzi P, Toto M, Margari L. A childhood case of symptomatic essential and psychogenic palatal tremor. Neuropsychiatr Dis Treat 2011;7:223-227.
3. Deuschl G, Toro C, Valls-Solé J, Zeffiro T, Zee DS, Hallet M. Symptomatic and essential palatal tremor. 1. Clinical, physiological and MRI analysis. Brain 1994;117(Pt 4):775-788.
4. Deuschl G, Toro C, Valls-Solé J, Hallet M. Symptomatic and essential palatal tremor. 3. Abnormal motor learning. J Neurol Neurosurg Psychiatry 1996;60:520-525.
5. Deuschl G, Toro C, Hallet M. Symptomatic and essential palatal tremor. 2. Differences of palatal movements. Mov Disord 1994;9:676-678.
6. Deuschl G, Wilms H. Clinical spectrum and physiology of palatal tremor. Mov Disord 2002;17 Suppl 2:S63-S66.
7. Deuschl G, Wilms H. Palatal tremor: the clinical spectrum and physiology of a rhythmic movement disorder. Adv Neurol 2002;89:115-130.
8. Zadikoff C, Lang AE, Klein C. The ‘essentials’ of essential palatal tremor: a reappraisal of the nosology. Brain 2006;129(Pt 4):832-840.
9. Stamelou M, Saifee TA, Edwards MJ, Bhatia KP. Psychogenic palatal tremor may be underrecognized: reappraisal of a large series of cases. Mov Disord 2012;27:1164-1168.
10. Gupta A, Lang AE. Psychogenic movement disorders. Curr Opin Neurol 2009;22:430-436.
11. Thomas M, Jankovic J. Psychogenic movement disorders: diagnosis and management. CNS Drugs 2004;18:437-452.