



Isolated lingual dystonia induced by speaking: a rare form of focal dystonia

Yildiz DEGIRMENCI, Ceyda Hayretdag ORS, Yeliz YILMAZ and Handan Isin Ozisik KARAMAN
Neurology Department, Canakkale Onsekiz Mart University Faculty of Medicine, Canakkale, Turkey

Abstract

Focal lingual dystonia is a rare condition that can be misdiagnosed as a psychogenic problem because it may interfere with chewing, swallowing, and speaking. We present a patient with an uncommon type of dystonia (speech-induced primary lingual dystonia), that responded well to botulinum toxin injection.

Key words: Lingual dystonia; treatment; Botulinum toxin injection.

Introduction

Lingual dystonia is a rare, disabling form of focal dystonia that impacts on daily activities including speaking, chewing, and swallowing, and causes social and vocational disabilities. The movements vary, from repetitive and/or episodic to sustained tongue protrusion, and can also be action induced with speaking or eating (1-3). Lingual protrusion often occurs in association with oromandibular dystonia (OMD), which can present as a symptom of varicella infection, orofacial injury or use of neuroleptic drugs (4-7). It can be isolated as well (8). However, primary lingual dystonia induced by speaking is a rare type of focal dystonia that is usually idiopathic in origin and is characterized by increased tonus of the tongue, which causes protrusion only during speaking. This report describes a rare condition of isolated lingual dystonia only induced by speaking.

Case report

A 42-year-old, right-handed woman presented to our outpatient clinic with a speech disturbance complaint that had begun 2-3 years ago. She was able to eat and drink normally. She had no family history of neurological disease and no history of neuroleptic drug use. Her neurological examination was normal

except for uncontrolled protrusion of her tongue during speech which was causing dysarthria. This abnormal movement was leading to unpredictable sounds when the tongue touched her front teeth. She felt more comfortable when she had something in her mouth, such as gum or candy. Brain magnetic resonance imaging and electroencephalography were normal. Standard blood tests, including tests for thyroid, parathyroid, and Wilson's disease were within normal limits. Psychiatric consultation was also normal except for depressive symptoms due to her presenting symptoms. She had a history of unsuccessful anticholinergic drug (trihexyphenidyl) treatment for approximately 4 months. EMG-guided 2X40 unit Botulinum toxin (Dysport®) injection was applied to the genioglossus muscle, and the patient was evaluated a month after injection.

Discussion

Focal lingual dystonia is a rare condition that can be misdiagnosed as a psychogenic problem because it may interfere with chewing, swallowing, and speaking. Our case had a lingual dystonia which was relieved by sensory tricks such as, when something was in her mouth. Some previous reports about speech induced focal dystonias described clinical signs extending to the orofacial area, not only the lingual area (8): some others did not mention sensory tricks (2). There was no psychological cause for our patient's lingual dystonia, no history of neuroleptic drug use or head trauma and there was no relevant family history. While prescription of anticholinergic medication is the most common treatment of lingual dystonia, our patient had a history of unsuccessful response to this treatment. Thus, our second option was to perform EMG-guided botulinum toxin injection into the dystonic muscle.

We present an uncommon type of dystonia (speech-induced primary lingual dystonia), which

may be difficult to differentiate from psychogenic dystonia. The lingual dystonia failed to respond to medical treatment, but responded well to botulinum toxin injection.

REFERENCES

1. Baik JS, Park JH, Kim JY. Primary lingual dystonia induced by speaking. *Mov Disord.* 2004;19:1251-2.
2. Ishii K, Tamaoka A, Shoji S. A case of primary focal lingual dystonia induced by speaking. *Eur J Neurol.* 2001;8:507.
3. Tan EK, Chan LL. Sensory tricks and treatment in primary lingual dystonia. *Mov Disord.* 2005;20:388.
4. Gollomp SM, Fahn S. Transient dystonia as a complication of varicella. *J Neurol Neurosurg Psychiatry.* 1987;50:1228-1229.
5. Ondo W. Lingual dystonia following electrical injury. *Mov Disord.* 1997;12:253.
6. Tan EK, Jankovic J. Tardive and idiopathic oromandibular dystonia: a clinical comparison. *J Neurol Neurosurg Psychiatry.* 2000;68:186-90.
7. Marsden CD. Blepharospasmeoromandibular dystonia syndrome (Brueghel's syndrome). A variant of adult-onset torsion dystonia? *J Neurol Neurosurg Psychiatry.* 1976;39:1204-9.
8. Paulson GW, Barnes J. Oral facial dystonia triggered by speech. *Psychosomatics.* 1988;29:236-238.

Dr. Yildiz Degirmenci,
Canakkale Onsekiz Mart University,
Neurology Department,
Canakkale (Turkey).
E-mail: ydegir@gmail.com