

# Speech-induced lingual dystonia

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Dystonia can be classified according to its etiology as primary, dystonia-plus, secondary, hereditodegenerative, and psychogenic<sup>1</sup>. Pure lingual dystonia is rare but not uncommon in association with other cranial dystonia. However, in clinical practice, even after excluding the main secondary causes of dystonia, the differential diagnosis between primary *versus* psychogenic forms may be tricky<sup>1</sup>. There are few cases in the literature describing speech-induced lingual dystonia and they were mostly classified as idiopathic<sup>2-5</sup>. We report a case of a woman who presented with a speech-induced lingual dystonia and discuss the differential diagnosis and potential therapeutic options of this condition.

## CASE

A 49-year-old white woman was referred to our service with a six-year history of tongue protrusion when speaking. There was no relevant past or family history of neurological disorders. She did not take neuroleptics or other medications before the onset of symptoms and had no history of facial injury or infection. For six years she visited several physicians and no diagnosis was made. On her first appointment at our service she was taking clonazepam 2 mg/day without improvement. Her neurological exam showed speech-induced tongue protrusion associated with mild dysarthria. The movement disorder showed no improvement with chewing gum (sensory trick). The patient could eat, drink, whistle, sing, and whisper without any trouble. A trial with levodopa (750 mg/day) and then trihexyphenidyl (10 mg/day) did not ameliorate symptoms.

A number of exams were ordered to rule out secondary dystonia. Drug-induced, dopa-responsive, post-traumatic and post-infectious dystonias had already been ruled out, and the absence of family history suggested no hereditodegenerative disorder. At this time the diagnoses considered were neuroacanthocytosis, Wilson's disease, and pantothenate kinase-associated neurodegeneration. Routine hematological and biochemical evaluation were completely normal including copper levels and number of acanthocytes. Brain magnetic resonance imaging (MRI), electroencephalography (EEG), and electromyography (EMG) were within normal parameters.

A speech-therapist evaluated the tongue movement during several tasks: repeated words and sentences, reading a short text, automatic speech, singing, vowel and fricative phoneme prolongation, sequences of syllables, and spontaneous conversation. The tongue movement disorder was identified in all circumstances of speech and in all phonemes, except vowel and sound prolongations. Tongue protrusion occurred more often in alveodental and alveolar phonemes and less frequently in palatal and velar phonemes. Slower speech and low voice intensity improved tongue protrusion.

After the initial work-up ruling out many etiologies, we investigated non-organic causes and referred her to a psychiatric examination. In this evaluation the patient told that her symptoms started during a period she went through a serious moral dilemma while working in an illegal informal job she considered humiliating. She also told that these symptoms could

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be related to psychological causes so that severity tended to increase when she was feeling herself guilty. During the psychiatric appointment the movement disorder improved with external suggestion that consisted of stopping tongue protrusion while speaking, achieving brief periods of full remission (lasting up to 5 minutes). The same improvement was not observed outside the medical office. According to the Structured Clinical Interview for DSM IV, she had the diagnosis of major depressive episode 8 years ago and no other diagnosis.

Although a psychogenic etiology for her dystonia could not be ruled out just because we did not identify sensory trick, positive neuroimaging or laboratory data, and positive neurophysiological studies, or due to the patient's personal problems and stressing events, we considered the etiology as primary. After a six-month follow-up, she still presents the same tongue movement disorder, although without specific medication. We did not use Botulinum Toxin (BTX) type-A injections since the movement disorder did not worsen. A signed-informed consent was obtained from the patient to allow her data publication.

### DISCUSSION

Tongue movement disorders comprise a poorly understood group of diseases, and several clinical presentations were described, such as tremor, myokimia and dyskinesias. However, there are few reports on speech-induced lingual dystonia<sup>2-5</sup>, and generally a primary etiology is considered.

Table shows a review of patients with speech-induced lingual dystonia. Two of the four cases reported in the literature considered a psychogenic etiology for lingual dystonia<sup>2,3</sup>. Interestingly, in both cases, interviews by psychologists and psychometric testing indicated a normal psychiatric state and ruled out a character disorder. In our case, we also considered a primary etiology despite several psychiatric stressors identified and negative work-up.

We made a thorough speech assessment of our patient. The goal was to evaluate any pattern of tongue protrusion dystonia, since no data about this issue was available in the literature. We found that the tongue movement disorder was present in all circumstances of speech and in all phonemes, except vowel and sound prolongations. Therefore, further studies comparing the pattern of speech abnormalities in patients with speech-induced lingual dystonia with healthy control subjects deliberately generating tongue protrusion during speaking would be of interest.

There are other unusual reports of perioral dystonia/dyskinesias affecting the lower facial muscles and jaw, but these disorders are mainly present at rest and seldom during speech<sup>6-8</sup>. Paroxysmal movement disorders such as paroxysmal kinesogenic dyskinesias and episodic ataxias are well described and usually involve the limbs.

Table. Literature findings on the reported patients with speech-induced lingual dystonia.

Case number	Age onset gender	Disease duration	Previous exposure neuroleptics / trauma / infections	Spreading dystonia-type	Sensory tricks	Etiology	Medication	Outcome	Reference/year
1	32 y-o Male	Weeks (?)	No	Yes (masseter) protrusion	No	Primary (psychogenic?)	Baclofen (?) Tizanidine (?) Trihexyphenidyl (8 mg/day)	Full recovery	Ishii, Tamaoka, Shoji, 2001 <sup>2</sup>
2	46 y-o Woman	7 Months	No	No protrusion	Yes	Primary (psychogenic?)	Trihexyphenidyl (4 mg/day)	Full recovery	Balk, Park, Kim, 2004 <sup>3</sup>
3	50 y-o Woman	Months (?)	No	No retraction	Yes	Primary	Anticholinergics (?) Tetrabenazine (?) Benzodiazepines (?)	Unchanged	Tan, Chan 2005 <sup>4</sup>
4	37 y-o Male	2 Years	No	No retraction	Yes	Primary	Trihexyphenidyl (10 mg/day)	Mild (~20%) improvement	Papapetropoulos, Singer 2005 <sup>5</sup>
5	49 y-o Woman	6 Years	No	No protrusion	Yes	Primary (Psychogenic?)	Clonazepam (2 mg) Levodopa (750 mg/day) Trihexyphenidyl (10 mg/day)	Unchanged	Felicio et al. 2009*

\*Present study.

Spasms of the tongue are seldom reported to occur in these syndromes<sup>6,7</sup>. Therefore, episodic or paroxysmal movement disorders involving the tongue although rare should be taken as differential diagnosis for patients with dystonia speech-induced. Focal lingual dystonia or tremor have been associated with trauma, especially with electrical injuries and is another form of exclusive involvement of the tongue<sup>9</sup>.

The best treatment for the primary forms of oro-buccal-lingual dystonias is BTX injections<sup>10</sup>. No patients reported with speech-induced lingual dystonia (including ours) were treated with BTX<sup>2-5</sup>. Actually, anticholinergic agents were the oral medications more often prescribed (Table).

Treatment with BTX type-A injection in the genio-glossus in a series of nine patients with involuntary tongue protrusion due to oromandibular dystonia or Meige's syndrome showed marked reduction in tongue protrusion in six patients, suggesting that BTX type-A may be a valid option for involuntary tongue protrusion<sup>11</sup>. Dysphagia is a frequent complication of treating lingual dystonia with BTX and it may lead to choking. Therefore, further studies are needed to better evaluate the efficacy of this BTX in lingual dystonia, minimizing dysphagia, an undesirable side-effect<sup>12</sup>.

In conclusion, speech-induced lingual dystonia should be considered as one of the presentation forms of task-specific tongue movement disorders and physicians should be aware of its differential diagnosis.

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