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LETTER TO THE EDITOR

Speech-Induced Task-Specific Cranio-Cervical Tardive Dystonia: An Unusual Phenomenology

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Dear Editor,

Dystonia is defined as sustained or intermittent muscle contractions causing abnormal, often repetitive, movements, postures, or both. Task-specific dystonia (TSD) encompasses a group of focal dystonias, affecting an isolated body part, predominantly laryngeal and hand muscles, and triggered by a specific and often skilled task. Frequently reported TSDs include laryngeal dystonia, writer's cramp, musician's dystonia, and occupational dystonia. Speech-induced dystonia often involves laryngeal and cranial muscles, with a speech-induced cervical TSD very rarely reported.[1,2] This report describes an unusual case of a speech-induced segmental TSD affecting the cranio-cervical area and probably having a drug-induced etiology.

A 29-year-old man presented with an eight-month history of progressive abnormal movements of the face and neck during vocalization. These movements were maximal during initiation of the speech, then reduced in a few seconds of continuing to speak and were absent at rest. There was no swallowing or breathing difficulty. The patient was borne to consanguineous parents and had a mild global developmental delay and a history of stammering in early childhood, which improved over time. He was on olanzapine (10 mg/d) and sodium valproate (500 mg/d) for the past year, which was started for episodic manic bipolar affective disorder (BPAD). Abnormal movements started 4 months after starting the treatment for BPAD. There was no significant family history. A video of the patient was recorded after obtaining written informed consent.

On examination (Supplementary Video 1, segment 1 in the online-only Data Supplement), there were mild right laterocollis and left torticollis at rest, with a normal strength and range of neck movements. Upon attempting to speak (Supplementary Video 1, segment 2 in the online-only Data Supplement), there were retrocollis and right torticollis, along with a forehead muscle contraction, widening of the palpebral fissure, and jaw opening, initially lasting for 2–3 seconds and returning to normal upon continued vocalization but recurring upon attempting to speak again. These movements predominantly occurred at the initiation of speech or phonation. They were also triggered by a silent speech, whispering, and singing, with the same severity but no language specificity. There were no sensory trick, voice tremor or abnormal neck movements during sustained phonation (Supplementary Video 1, segment 3 in the online-only Data Supplement). Dystonia was not triggered by protruding the tongue, opening the mouth, showing the teeth, voluntarily closing the eyes, swallowing, gulping or blowing air. Additionally, there was a postural lingual dystonia and mild postural distal hand dystonia, unrelated to speech. The rest of his neurological and other systemic tests were normal.

Routine blood tests, serum and urine copper tests, and magnetic resonance imaging of the brain were normal, and a slit-lamp examination for a Keyser-Fleischer ring was negative. The patient was managed as a case of speech-induced segmental TSD affecting the cranio-cervical area (Axis-I), with a probable drug-induced or idiopathic etiology. In the absence of an urge, suppressibility, and a rebound phenomenon, a complex motor tic seemed unlikely. There was no distractibility, entrainability or inconsistency in movements to suggest a functional movement disorder. Dystonia improved by approximately 90% after stopping olanzapine and starting tetrabenazine (25 mg/d). The valproate dose was adjusted for the management of BPAD. Dystonia did not

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recur after stopping tetrabenazine during a 3-month follow-up.

To the best of our knowledge, only two cases of speech-induced cervical TSD have been reported so far in the literature.[1,2] Both cases only had cervical dystonia, unlike our case who also had cranial involvement. The etiology was probably idiopathic in both cases, as the workup for secondary causes was negative, and there was no history of prior drug intake. In our case, there was a history of exposure to atypical antipsychotics and improvement upon withdrawing an antipsychotic, suggesting a diagnosis of tardive dystonia. In addition, retrocollis is more frequently observed in tardive than in idiopathic cervical dystonia. There are previous reports of TSDs after neuroleptic use, which were predominantly oromandibular and lingual dystonias, unlike that in our patient, which also included the cervical region. Two cases of a cervical TSD triggered by tasks other than speech have also been reported. In one patient, TSD was triggered by cradling the phone between the head and shoulder, and in the other, who was a bilateral traumatic arm amputee, it was triggered by writing with a pen held in the mouth.[3,4]

The etiology of most TSDs remains unclear and can be polygenetic or environmental.[5] Impaired inhibition and abnormal, excessive, topographically unspecific neuronal plasticity are two dominant hypotheses put forth to explain the task specificity.[5] A complex interplay among genetic predisposition, drug overuse, and biochemical and psychological factors appears to underlie impaired inhibition and plasticity. These factors may lead to abnormal reorganization of somatotopic representations within sensorimotor regions in these patients. However, in tardive syndromes, dopaminergic hypersensitivity, an imbalance between dopamine and cholinergic systems, strio-nigral GABAergic neuronal dysfunction, excitotoxicity and oxidative stress, leading to structural abnormalities, have been implicated.[6] Additionally, genetic vulnerability has been postulated in both tardive syndromes and TSD. The consanguineous parentage, developmental delay and stammering in early childhood may suggest an underlying genetic predisposition in our patient, which may have become unmasked by the use of neuroleptics for his BPAD, resulting in tardive TSD. However, a possibility of a pure idiopathic or underlying genetic cause cannot be entirely ruled out as we could not perform genetic assessment.

Speech-induced dystonia is a rare form of TSD, usually causing oral, lingual, labial or mandibular dystonia, in isolation or in various combinations.[2] This form can rarely be contextual, e.g., while praying, auctioneering or reciting mantras.[7] It may remain task specific or, over time, can be triggered by other tasks. Speech-induced dystonia can evolve into persistent dystonia and spread to adjacent body regions as well. It is common that the muscles involved in a respective task are involved first, with dystonia later spreading to other surrounding muscles. However, in our case, even though there was a lingual and jaw dystonia, there was no significant vocal cord involvement, and cervical dystonia was more prominent. The exact pathophysiology of speech-induced dystonia is unclear. Speech inducibility may suggest abnormal activation and dystonic overflow to the surrounding facial, lingual, labial, mandibular and cervical musculature, with laryngeal activation due to abnormal surrounding inhibition in the cortex.[2]

In conclusion, even though speech-induced dystonia commonly involves craniofacial muscles, it can rarely lead to a task-specific cranio-cervical dystonia. Tardive dystonia should be considered while evaluating the etiology of such cases.

Supplementary Video Legends

Video 1.

Segment 1: video of the patient, showing mild right lateral collis and left torticollis at rest, with lingual dystonia upon tongue protrusion. There was no dystonia when the patient was asked to close his eyes.

Segment 2: cranio-cervical dystonia triggered by speech when attempting to count numbers and tell the name of the month.

Segment 3: dystonia subsides after an initial 2−3 seconds of sustained phonation.

Supplementary Materials

The online-only Data Supplement is available with this article at https://doi.org/10.14802/jmd.20067.

Conflicts of Interest

The author has no financial conflicts of interest.

Acknowledgments

None.

Ethical Standard

Ethical approval was waived by the Institutional Review Board. The video of the patient was recorded after obtaining written informed consent. I confirm that I have read the Journal's position on issues involved in ethical publication and affirm that this work is consistent with those guidelines.

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