Speech-Induced Cervical Dystonia

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Dystonia is characterized by sustained involuntary muscle contractions leading to repetitive twisting movements or abnormal postures. Such abnormal muscle contractions can be focal (affecting only one body part, such as blepharospasm), segmental (affecting at least two adjacent areas of the body such as Meige syndrome), multifocal (involving two or more non-adjacent areas of the body) or generalized (involving at least one leg and the trunk as well as one other area of the body). Dystonia can be spontaneous and permanent or triggered by the performance of a specific action, then called "task-specific dystonia" (TSD). The most common forms of TSD are writer's cramp, musician's cramp, embouchure dystonia, sports-related dystonia (golf, darts) and occupational dystonia (typist, telegraphist). Task-specific dystonia typically affects the hand, or occasionally the face, following the performance of a stereotyped, repetitive highly skilled movement. This article describes the case of a patient with an unusual form of task-specific cervical dystonia, which is speech induced. Speech-induced cervical dystonia is uncommon and has never been described previously.

Case report

A 20 year-old man from Pakistan is referred to our movement disorders clinic for a five year history of progressive abnormal neck movements associated with speech. On initiation of speech, the patient's neck flexes and tilts to the right. Resisting neck movement prevents speech production. Neck movements do not occur with laughing, whispering or singing, and are never present without speech. The patient does not describe any urge associated with neck movements and the contractions are not suppressible. He denies any other abnormal movements, tics or dysphagia. Past medical history is unremarkable. There is no past history of exposure to dopamine blocking agents.

On examination, there is no deviation of the head or abnormal contraction of neck muscles when the patient is silent. Neck movements and strength are normal. Repetitive head flexion with mild to moderate right laterocollis occurs only during speech (see video on-line). When asked to sustain a vowel or consonant, neck contraction occurs at speech onset only. Mild dysphonia is also present at speech onset. Speaking his native language does not change the head movements, but they disappear with whispering. Palpation of his neck during speech reveals contraction of both sternocleidomastoid muscles as well as lesser platysma and scalene involvement. The rest of neurological examination is unremarkable. Thyroid function, ceruloplasmin, 24-hour urine copper, brain and cervical-spine magnetic resonance imaging are normal.

Discussion

Task-specific dystonia consists of abnormal muscle contractions or abnormal postures occuring only while performing a specific motor task. Common TSD include involuntary contraction of the hand or forearm muscles when writing (writer's cramp), abnormal contraction of the mouth or hand muscles when playing a musical instrument (embouchure dystonia or musician's cramp) and a variety of occupation-related or sports-related dystonia such as involuntary cramping of the hand when typing, using a mouse, golfing, pistol-shooting, playing tennis table, etc[1-4]. The trigger action, in most cases, is a highly skilled, stereotyped, overlearned motor task[1,5,6]. Over time, a TSD can spread to other tasks or evolve into a countinuously present dystonia, possibly spreading to adjacent body regions[1,2,7]. The pathophysiology of TSD is uncertain, but is thought to relate to dysfunction of brain plasticity leading to loss of inhibition in the motor system at various levels, reduced sensory perception and integration, and impaired sensorimotor processing and integration[1,2,6]. However, most studies concerned writer's cramp and musician's dystonia and it is unclear whether the results can be applied to other forms of TSD. Different anatomic sites, demographics of affected individuals and prognosis indicate that all patients may not share the same pathophysiology[1].

Only one case of task-specific cervical dystonia has been described[7]. The reported patient had suffered bilateral traumatic arm amputation and learned to write and draw using tools held in his mouth. He developed cervical dystonia triggered by writing and drawing, which eventually evolved into a constant cervical dystonia[7]. It is interesting to note that in this case, a TSD appeared when the neck became involved in a highly skilled and repetitive motor task.

Our case presents with unsupressible head movements triggered by speech, thus diagnosed as speech-induced cervical dystonia, a form of TSD. The absence of head movement with laughing or whispering supports our diagnosis of task-specific dystonia. A diagnosis of motor tic was excluded given the absence of a premonitory urge and the absence of supressibility. A psychogenic movement disorder was considered less likely given the consistency of the abnormal movements (even when

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the patient was not aware of being watched), consistency of physical examination, absence of distractibility, absence of psychiatric comorbidites and absence of pending litigation.

Speech-induced dystonia is a rare form of TSD, usually affecting the facial musculature. Involuntary muscle contractions can result from any speech output, as seen in our case, or can be restricted to a specific content such as recitation of a Buddhist mantra, prayers in a certain language or auctioneering[4]. Previously published cases involved speech-induced blepharospasm, dystonia of the lip, lower face, jaw or tongue[4,8]. It has been suggested that speech inducibility in such cases reflects a dystonic overflow to facial muscles with activation of the larynx due to abnormal surrounding inhibition in the cortex[8]. A similar pathophysiology could apply to our case, with adequate inhibition of the facial muscles but a a dystonic overflow spreading to the cervical muscles.

In conclusion, TSD is a form of dystonia triggered by a specific action, commonly a highly skilled and repetitive task. Most common TSD involve the hands or the face and are associated with writing, playing a musical instrument, practising a sport or a job-related task. Task-specific dystonia involving the neck is extremely rare. Dystonia induced by speech is also rare, and usually involves facial muscles. The combination of speech-induced cervical dystonia is therefore unusual, although it may share a similar pathophysiology as other forms of TSD, where a dysfunction of brain plasticity and loss of inhibition in the motor system plays a central role.

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