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Botulinum toxin in a task-specific oromandibular dystonia in a bingo caller

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Sirs: Focal task-specific dystonia is a subset of action dystonia that affects one part of the body, and occurs exclusively during the performance of a specific act. This disorder generally affects the hands when writing, typing, playing musical instruments. In contrast, there have been few descriptions of task-specific oromandibular dystonia (OMD), mainly in wind instruments players. We present a case of occupational OMD in a bingo caller, whose perioral dystonic movements interfered with her pronunciation of the bingo numbers.

A 47-year-old female presented to our department with an 11 month history of progressive difficulty in calling the bingo numbers as a result of involuntary oromandibular movements while performing this activity. This had been her job for the previous 5 years. At that moment, her speech was unaffected during conversation and had no difficulty in chewing and swallowing. She did not refer other symptoms.

She had no familiar history of movement disorders, nor relevant

past medical history, nor exposure to neuroleptics, and no recent oromandibular trauma.

The general physical and neurological examinations were both normal, apart from the task-specific OMD. A dystonic contraction of perioral muscles, with protrusion of the tongue, retraction of the jaw, and dysarthria, could only be provoked by persuading the patient to call the bingo numbers. This condition disappeared at rest, and recurred when recommencing this activity.

The complete blood tests, including examinations for acanthocytes, thyroid function, microbiologic study, and copper metabolism, were normal. Cranial magnetic resonance imaging and electromyography of the lower facial muscles showed no abnormalities.

Her OMD continued worsening gradually over the next few months and the consequent slurring forced her to retire from her profession. One year later, she began to experience difficulties with ordinary speech. She was treated with clonazepam, tetrabenazine and carbamazepine, with no benefit. Then, we injected 75 units of botulinum toxin A (Dysport) into both medial pterygoid muscles, with a mild improvement. Four months later, a second infiltration of 125 units of botulinum toxin A into these muscles was dramatically effective. This good response was maintained on follow-up, with an infiltration every six months. The patient was able to return to her occupation.

Dystonic movements can occur at rest, but usually become pronounced by voluntary motor activity (action dystonia). One form of action dystonia is the occupational or task-specific focal dystonia present only during specific activity [3]. It generally involves the hands and affects individuals who per-

form complex repetitive movements. Writer's and typist's cramps are the most common. Other occupational focal dystonias affect musicians (pianist's and violinist's hand) [7, 9], sawyers, hammers, golfers [6], etc. Task-specific OMD appears less frequently and most cases have been described in wind instruments players [1]. Only two cases of occupational OMD involving a specific use of speech have been previously reported, affecting an auctioneer [8] and an Islamic prayer [2]. We present another case of this unusual task-specific dystonia in a bingo caller.

The response of OMD to pharmacotherapy including anticholinergic agents, dopamine depleting agents, or GABA agonists is frequently unsatisfactory. Although the botulinum toxin is recommended as a suitable therapy for OMD [4], the improvement appears to be lower in comparison to other dystonias [5]. In a report of embouchure dystonia [1], only one out of the seven woodwind players who underwent botulinum toxin treatment experienced a significant benefit. Nevertheless, the botulinum toxin injections were effective in our patient and in the auctioneer's dystonia case [8].

This case illustrates an unusual occupational OMD that exclusively appeared in a specific use of speech, with later spreading to other oral task. Treatment is often difficult. In our patient, botulinum toxin injections provided a dramatic improvement. We consider this treatment as a therapeutic option for task-specific OMD.

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