

## Chewing-induced Facial Dystonia

### Dear Editor,

Dystonia is a movement disorder characterised by sustained muscle contractions, repetitive twisting movements, and abnormal postures of the trunk, neck, face, or arms and legs.<sup>1</sup> Classification of this condition may be based on topography, age at onset, cause, or by the presence of genetic markers.<sup>1</sup> When unaccompanied by other neurologic abnormalities, and in the absence of known causes (except some genetic mutations), the dystonia is said to be “primary”. Focal dystonia is that which affects a single region, e.g. cervical dystonia, blepharospasm or laryngeal dystonia. Dystonia may also be classified according to phenomenology. Hence, abnormal posturing that occurs during voluntary activity is called “action dystonia”, the eponym “task-specific” being appended when action dystonia is triggered by specific actions.<sup>1</sup>

### Case Report

A previously well 37-year-old man presented to our clinic with the complaint that, for the preceding 6 months, he “made faces” when eating large mouthfuls of food, but not otherwise. The movements, which were likened to the “face made when confronted with a noxious odour”, were consistent and reproducible, occurring only when he chewed large mouthfuls of food, but not when he chewed small mouthfuls of food, talked, sang or performed other tasks. They caused the patient severe distress and social embarrassment, and were upsetting to his wife, who initially misinterpreted his facial expression as indicating lack of appreciation of her cooking. He had no previous history of psychiatric illness or head injury, had no history of facial palsy, and had never ingested antipsychotic agents. There was no family history of movement disorders. The abnormal movements were not preceded by an urge to move, and their performance did not provide relief. He did not describe any sensory tricks which ameliorated the movements, and had no brady- or hypokinesia, rest tremors or stiffness to suggest a parkinsonian syndrome.

Clinical examination was normal, except for overactivity of the procerus, corrugator and nasalis muscles, causing him to produce the “bad smell facies” when he chewed on a large mouthful of food. These abnormal movements were not otherwise seen, even when he drank or chewed

on smaller mouthfuls of food or mimicked chewing, without food in the mouth. He did not adopt any abnormal posturing when writing, did not blink excessively or forcibly close his eyes, tilt his head or speak with a strangulated or breathy tone. Neuroimaging and blood tests, including thyroid function, peripheral blood film and Wilson’s disease work-up, were normal. He was diagnosed to have task-specific facial dystonia, triggered by chewing, and elected to receive injections of botulinum toxin, BTX (100 units of Botox®, Allergan, Irvine, CA, reconstituted in 2.0 mL of 0.9% normal saline at a concentration of 5 units/0.1 mL) totaling 35 units to the above muscles (7.5 units to each corrugator, 5 units to each side of the procerus and nasalis muscles). Within 2 weeks, the abnormal facial movements abated, returning only after a period of 4 months. He had no facial weakness resulting from the injections, and reported complete satisfaction with the injections, choosing to repeat the injections at 4-monthly intervals.

### Discussion

Although action-induced hemifacial spasm has recently been described,<sup>2</sup> the involvement of the procerus, corrugator and nasalis rather than the orbicularis oculi, bilateral facial involvement and task-specificity favoured a diagnosis of focal task-specific dystonia (FTSD) rather than hemifacial spasm.<sup>3</sup> The diagnosis of motor tics was likewise excluded because of the task-specificity, confinement of abnormal movements to the upper face, absence of premonitory symptoms, lack of sensation of relief after twitching and the sustained, rather than rapid, movements.<sup>4</sup> Limitation of twitching to the face and lack of exposure to neuroleptic agents likewise excluded a diagnosis of tardive dyskinesia.<sup>5</sup> The consistency of the orofacial movements, lack of distractibility or entrainment and absence of psychiatric history or psychoneurotic features excluded a psychogenic movement disorder,<sup>6</sup> although it was not possible to observe him when unaware of our presence. Although the “procerus sign” has been described in progressive supranuclear palsy and corticobasal degeneration,<sup>7</sup> the absence of typical features of bradykinesia, cognitive impairment, stiffness and limitation of extraocular movements at first consultation and during subsequent clinic visits over the next 3 years, excludes a parkinsonian syndrome. Finally, a diagnosis

of synkinesis, i.e. inappropriate contraction of muscles innervated by the facial nerve with activation of muscles supplied by the trigeminal nerve, secondary to aberrant regeneration of a damaged facial nerve,<sup>2</sup> was excluded by the normal MRI brain and absence of past history of facial palsy.

Focal task-specific dystonias usually involve the upper limbs or craniocervical region, although task-specific dystonia of the lower limbs has recently been described.<sup>8</sup> Furthermore, they are usually associated with stereotyped, highly repetitive movements. Musician's and writer's cramps are FTSDs of the arm and hand muscles, which manifest when playing musical instruments or writing. Craniocervical dystonia<sup>9</sup> and purely cranial dystonias, such as embouchure dystonia of the orobuccal muscles (observed in woodwind and brass musicians), as well as oromandibular dystonia, occurring in bingo callers, auctioneers and in religious personnel reciting mantras and Islamic prayers, have also been described.<sup>10</sup> Whether our patient had FTSD, or focal dystonia coincidentally triggered by chewing is debatable, especially since the dystonia occurred whilst eating large boluses of food, an act which is neither highly stereotyped nor frequently repeated. Lagueny et al<sup>11</sup> described a similar case of FTSD of the jaw, occurring only when chewing food of hard consistency, and postulated that the FTSD was triggered by pressure on the periodontal mechanoreceptors, and a defective central control of their sensory inputs. Other mechanisms may be considered to explain the chewing-induced facial movements. An artificial synapse, i.e. ephaptic transmission between the facial and trigeminal nerves (which lie in close proximity, especially at their point of origin in the pons), may be invoked.<sup>12</sup> The phenomenology of our patient's chewing-induced facial dystonia is consistent with FTSD.

## REFERENCES

1. Tarsy D, Simon DK. Dystonia. *N Engl J Med* 2006;355:818-29.
2. Yang SS, Seet RC, Lim EC. Action-induced hemifacial spasm and its resolution with botulinum toxin. *Mov Disord* 2009;24:147-8.
3. Tan EK, Jankovic J. Bilateral hemifacial spasm: a report of five cases and a literature review. *Mov Disord* 1999;14:345-9.
4. Kwak CH, Hanna PA, Jankovic J. Botulinum toxin in the treatment of tics. *Arch Neurol* 2000;57:1190-3.
5. Lim EC, Wilder-Smith EP, Seet RC. A blessing in disguise: resolution of tardive dyskinesia with development of cervical myelitis. *Mov Disord* 2006;21:120-2.
6. Schrag A, Lang AE. Psychogenic movement disorders. *Curr Opin Neurol* 2005;18:399-404.
7. Morimatsu M. Procerus sign in progressive supranuclear palsy and corticobasal degeneration. *Intern Med* 2002;41:1101-2.
8. Lo SE, Frucht SJ. Is focal task-specific dystonia limited to the hand and face? *Mov Disord* 2007;22:1009-11.
9. Schramm A, Neumann M, Reiners K, Classen J. Task-specific craniocervical dystonia. *Mov Disord* 2008;23:1041-3.
10. Pek CH, Seet RC, Yik JH, Lim EC. Orofacial dystonia triggered by mouth closure. *Clin Neurol Neurosurg* 2010;112:79-81.
11. Lagueny A, Caix P, Schuermans P, Julien J. Jaw dystonia triggered by biting into hard food. *Mov Disord* 1991;6:174-6.
12. Nielsen VK. Electrophysiology of the facial nerve in hemifacial spasm: ectopic/ephaptic excitation. *Muscle Nerve* 1985;8:545-55.

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