CASE REPORT

Chewing-induced facial dystonia: a rare presentation of task-specific dystonia

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SUMMARY

This case is an addition to scarce literature available for a rare condition, chewing-induced task-specific dystonia. The patient was a 63-year-old woman who presented with a 4-year history of progressive difficulty in eating food only during chewing associated with abnormal facial grimaces without any difficulty in drinking, swallowing, speaking or singing. Examination revealed dystonia of facial muscles every time she chewed but absent during drinking and speaking. As movements were consistent and reproducible with the specific task, other differential diagnosis like motor tics, psychogenic disorder, tardive dystonia and parkinsonism syndrome were excluded leading to a diagnosis of task-specific facial dystonia triggered by chewing. Treatment was started with trihexyphenidyl and later on tetrabenazine was also added but she got only mild relief of symptoms. As she did not agreed for botulinum toxin therapy, so we continued with the same treatment without much improvement.

BACKGROUND

Task-specific dystonia is primary focal dystonia characterised by excessive muscle contractions producing abnormal postures during selective motor activities that often involve highly skilled, repetitive movements.[1]

Despite the fact that first descriptions of task-specific dystonia (writer's cramp) was given by Bernardino in 1713 in his book of occupational diseases,[2] and in 1982, works of Sheehy and Marsden disproved the psychogenic nature of task-specific dystonias,[3] still only a handful of case reports are there for chewing-induced facial dystonia,[4–6] and our case is an addition to that and can give better insight into clinical features of this disorder.

CASE PRESENTATION

A 63-year-old woman presented with a 4-year history of progressive difficulty in chewing. Whenever she puts food bolus in her mouth she was not able to chew it properly as if both jaws are not coming in contact properly with each other and take abnormally longer time to chew than before and also bite her tongue and inner cheeks repeatedly. However, she had no difficulty in swallowing food, drinking, speaking or singing bhajans. Her relatives noted that she made abnormal grimaces while chewing the food, as if she does not like the taste of food and due to this patient has severe mental distress and social embarrassment. There is no history of fatigability or diurnal variation of symptoms.

There was no history of excessive or prolonged chewing of tobacco, betel nuts or chewing gum and no history of abnormal posturing in any other parts of the body. There was no inner urge prior to and relief after performing movements. She did not describe any sensory trick to abolish them. Also, there is no history suggestive of slowness in daily routine activities or rest tremor or stiffness. Her medical history was unremarkable for jaundice, head injury, facial palsy, psychiatric illness and ingestion of any dopamine-blocking agents including antipsychotics or antiemetics and there was no family history of abnormal movements.

On clinical examination, there were no abnormal movements at rest but when she was offered a biscuit to eat, there was abnormal contraction of the orbicularis oris, risorius, mentalis, procerus and frontalis muscles. These movements were consistent and reproducible every time she eats, but conspicuously absent when she drank or spoke. There were no abnormal movements elsewhere in the body, no finding suggestive of parkinsonism and no Kayser-Fleischer ring (K-F ring) seen on naked eye. Other neurological and other system examinations were also normal. These abnormal movements during chewing (online supplementary video 1) and the absence of it while speaking (online supplementary video 2) were recorded for future documentation.

INVESTIGATIONS

Workup for Wilson's disease including slit-lamp examination for K-F ring, 24-hour urinary copper and serum ceruloplasmin were within normal limits. MRI brain and other haematological and biochemical tests including thyroid function and liver profile were also normal.

DIFFERENTIAL DIAGNOSIS

Tardive dystonia was excluded due to the lack of exposure to dopamine- blocking agents.

Motor tics excluded by task specificity and due to the absence of premonitory symptoms.

Synkinesia after facial palsy was excluded due to no history of facial palsy.

Psychogenic movement disorder was excluded due to consistency of movements, lack of distractibility and no psychoneurotic features.

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Parkinsonism syndrome was excluded due to the absence of bradykinesia, rigidity, tremors or any other suggestive features.

Finally in view of treatable movement disorder, Wilson disease was ruled out due to the absence of K-F ring and negative investigations.

TREATMENT

She was started on tablet trihexyphenidyl 2 mg two times per day initially and then three times per day after 1 week.

Later on, tablet tetrabenazine 25 mg was also added; after 1 month of treatment, she was started on half a tablet two times per day and increased up to one full tablet three times per day over a period of 2 months.

OUTCOME AND FOLLOW-UP

She had only mild relief in her symptoms after 3 months of follow-up and planned for botulinum toxin therapy but she denied it.

DISCUSSION

Only handful of cases are reported so far as chewing-induced task-specific dystonia.[4–6] Yang *et al*[4] described a similar case of focal task-specific dystonia of face induced by chewing in which facial dystonia appear only on taking large boluses of food and had satisfactory recovery with botulinum toxin injections, while in our case dystonia appears with each bolus of food and she did not agree for botulinum toxin injection.

Bhattacharya *et al*[5] also described a similar case of chewing-induced dystonia of facial muscles.

Learning points

- Task-specific chewing dystonia is a rare entity.
- These dystonias can cause significant social embarrassment to patient and family.
- Botulinum toxin injection is the treatment of choice.
- High index of suspicion is required for correct diagnosis.

Yoo *et al*[6] also described oromandibular dystonia appearing only during the act of chewing in a 65-year-old woman which is phenomenological similar to our case but authors did not specified weather dystonia also involved facial muscles as in our case.

Lagueny *et al*[7] also described a similar case of dystonia of the lower jaw occurring while biting only hard food and also postulated that this is triggered by activation of periodontal mechanoreceptors and central control of their sensory inputs are defective. In the same way, prolonged chewing can induce oromandibular dystonia, as reported in patients from Middle East who used to chew khat (*Catha edulis*), a recreational substance, for a prolonged period prior to onset of dystonia.[8 9] Wadia and Khanna[8] proposed mechanical effects of prolonged chewing of khat as one of the aetiological factor but chemical effects from amphetamine-containing compounds, released during chewing of khat, may also play a role in pathophysiology as proposed by Shehata *et al*.[9]

Contributors All authors: manuscript preparation and reviewed the manuscript. AG: prepared and edited the manuscript.

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