



## Oromandibular Dystonia: A Rare Cause of Jaw Pain & Involuntary Jaw Movements- A Case Report

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### Abstract

*Oromandibular dystonia consists of sustained or intermittent involuntary, repetitive, patterned muscle contractions affecting the lower part of the face, mouth, jaw, tongue, and pharynx. Dystonia causes immense psychological distress and interferes with chewing, speech, and other functions. Although the condition is rare with prevalence of 3.4 per million, it is often misdiagnosed or left undiagnosed due to several patient-clinician factors.*

**Keywords-** *Oromandibular dystonia (OMD), involuntary spasmodic contractions, BoNT injections, EMG.*

### Introduction

Oromandibular dystonia (OMD) is a form of focal dystonia, characterized by involuntary repetitive movements of the jaw and forceful contractions of the face.<sup>1</sup> Dystonia causes immense psychological distress and interferes with chewing, speech, and other functions.<sup>2</sup> Based on the affected area, dystonia can be anatomically categorized as focal (affecting one or two parts of the body), segmental, multifocal, and generalized. It can also be categorized based on aetiology. Primary dystonia is idiopathic or inherited; while, secondary dystonia advances after traumatic or surgical incidents, brain diseases, and medications.<sup>1</sup>

Other terms for OMD are orofacial mandibular dystonia, orofacial buccal dystonia, lingual dystonia, jaw dystonia, cranial dystonia, and adult-onset facial dystonia.<sup>1</sup>

### Case Report

A 60-year-old female presented to department of oral medicine and radiology at College of Dental Sciences & Research Centre, Bopal, Ahmedabad with spasm of face and difficulty in speaking for 2 weeks. Patient was a known hypertensive for the past one year and was under regular medication for the same. On examination masseter and temporalis were tender (Gr-2 Acc. to J.J. Cipriano). She was not able to speak and eat properly due to pain (VAS 7). On intraoral examination her upper and lower posteriors were missing since last 2 months. Considering clinical presentation, a provisional diagnosis of myalgia was given. The patient was instructed to apply hot and cold fomentation, adhere to a soft diet, and limit mouth opening. A follow-up appointment was scheduled one week later, coinciding with the

appointment for prosthetic fittings in both the upper and lower regions.

On next appointment, pain was same and she had started experiencing spontaneous, intermittent, severely painful involuntary spasmodic contractions on the lower half of face involving tongue, lips which was continuous, repetitive throughout the day and which was not relieved on either on opening or closing of mouth to reappear again on next occlusal contact. During contraction tongue deviated, with slurring of speech.

Tardive dyskinesia, Oromandibular dystonia, Myokymia, Tourette syndrome was considered in the differential diagnosis. For ruling out the possibility patient was referred to neurophysician. Reassurance was the primary approach towards the treatment goal with a positive reinforcement of the curability of the disease. Patient was advised to undergo routine blood investigations along with MRI of brain. Her blood investigations were within normal limit and MRI showed mild cerebellar atrophy. Diagnosis of Oromandibular Dystonia was made and patient was prescribed Tablet Bexol (Trihexyphenidyl 2 mg) along with Tablet Baclof (Baclofen muscle relaxant 10mg) for 15 days. On 2<sup>nd</sup> follow-up visit Tablet Revocon 25 (Tetrabenazine) and Tab Clonotril (Clonazepam 0.25mg) was added alongside with prosthetic treatment.

After a month, she showed a definite reduction of the dystonic movement her spasmodic contractions had decreased although neurophysician had taught her a sensory trick to keep finger near her lip & chin region so that spasmodic contractions would decrease. After that patient was comfortable and prosthetic treatment was also completed. Patient is on continued treatment for the same.



**Figure 1:** Sensory Trick

### Discussion

Dystonia is defined as a movement disorder characterized by sustained or intermittent muscle contractions causing abnormal, often repetitive, movements, postures, or both. Dystonic movements are typically patterned, twisting, and may be tremulous, are often initiated or worsened by voluntary action and are associated with overflow muscle activation.<sup>3</sup> OMD consists of sustained or intermittent involuntary, repetitive, patterned muscle contractions affecting the lower part of the face, mouth, jaw, tongue, and pharynx.<sup>4</sup> It can be associated with difficulties in speaking, chewing, swallowing, and even breathing. Usual triggers include stress, speaking and swallowing. Prolonged jaw opening results in difficulty with mastication, swallowing and causes drooling. Patients have difficulty articulating and may have unintelligible speech. Jaw closing dystonia can occur alone, or in association with jaw opening dystonia.

The overall prevalence of primary dystonia is estimated as 164.3 per million. The prevalence of oromandibular dystonia is estimated to be around 68.9 per million.<sup>5</sup> The etiology of dystonia is multifactorial (neurological disorder, antipsychotic medications, hypoxic brain injury). Another method for classifying dystonia is primary (idiopathic, inherited or familial) or secondary.<sup>6</sup> It has been suspected that dental causes can be etiological factor for dystonia. In the present case patient was having missing teeth in upper and lower arch which could be the possible cause. OMD has no clinical diagnostic test hence, the diagnosis is based on history, neurological examination and confirmation by

intramuscular electromyography (EMG).<sup>1</sup> Multiple treatments are available for OMD such as pharmacological, sensory tricks and Botulinum toxin. Various agents utilized in the management of OMD include anticholinergics, baclofen, benzodiazepines, antiparkinsonian medications, anticonvulsants, carbamazepine, dopamine receptor antagonists, levodopa, and lithium.<sup>6</sup> Botulinum toxin a potent neurotoxin is a promising therapy for OMD.<sup>8</sup> BoNT injections can leave even better effects if guided with EMG, which allows longer intervals between the BoNT injections. It can cause hypo excitability of affected muscles. Various treatment modalities related to dental rehabilitation, such as acrylic replicas of sensory aids, implant-supported dentures, and removable appliances, have been proposed, each demonstrating a diverse range of treatment outcomes as reported in the literature.<sup>5</sup>

### Conclusion

Oromandibular Dystonia (OMD), which, although rare, can severely impact daily function and wellbeing. A multidisciplinary approach involving dental professionals and neurologists can be beneficial in managing such conditions. Since the OMD patients may present to dentists with involuntary jaw movements and intraoral finding/problems, the dentists should be aware of the symptoms and signs to refer the suspicious patients if necessary.

### References

1. Raoofi S, Khorshidi H, Najafi M. Etiology, Diagnosis and Management of Oromandibular Dystonia: an Update for Stomatologists. *J Dent (Shiraz)*. 2017 Jun;18(2):73-81.
2. Prabhakar V. Oromandibular Dystonia – Diagnosis not to be missed: A case report & review. *J Indian Prosthodont Soc*. 2020 Dec;20(Suppl 1):S28.
3. Albanese A, Bhatia K, Bressman SB, Delong MR, Fahn S, Fung VS, Hallett M, Jankovic J, Jinnah HA, Klein C, Lang AE, Mink JW, Teller JK. Phenomenology and classification of dystonia: a consensus update. *Mov Disord*. 2013 Jun 15;28(7):863-73.
4. D. Britton, J.E. Alty, C.J. Mannion, Oromandibular dystonia: a diagnosis not to miss, *British Journal of Oral and Maxillofacial Surgery*, Volume 58, Issue 5, 2020.
5. Yoshida K. Clinical and Phenomenological Characteristics of Patients with Task-Specific Lingual Dystonia: Possible Association with Occupation. *Front Neurol*. 2017 Dec 11;8:649. doi: 10.3389/fneur.2017.00649. PMID: 29321757; PMCID: PMC5732148.
6. Watt E, Sangani I, Crawford F, Gillgrass T. The role of a dentist in managing patients with dystonia. *Dent Update*. 2013 Dec;40(10):846-8. doi: 10.12968/denu.2013.40.10.846. PMID: 24597030.
7. Bledsoe IO, Viser AC, San Luciano M. Treatment of Dystonia: Medications, Neurotoxins, Neuromodulation, and Rehabilitation. *Neurotherapeutics*. 2020 Oct;17(4):1622-1644. Epub 2020 Oct 23. PMID: 33095402; PMCID: PMC7851280.
8. Karp BI, Alter K. Botulinum Toxin Treatment of Blepharospasm, Orofacial/Oromandibular Dystonia, and Hemifacial Spasm. *Semin Neurol*. 2016 Feb;36(1):84-91. Epub 2016 Feb 11. PMID: 26866500.