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Case Report

Management of Oromandibular Dystonia: A Case Report

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Abstract

Background: Oromandibular dystonia (OMD) is a rare focal dystonia involving involuntary contractions of masticatory, lingual, and facial muscles, leading to impaired speech, mastication, and swallowing. Its diagnosis is often delayed, and misinterpretation as psychogenic or temporomandibular joint disorders is common.

Case Presentation: We report the case of a 52-year-old woman with a history of depressive disorder, who presented with a two-year history of painful involuntary jaw-closing contractions, trismus, and dysarthria, severely impairing nutrition and daily life. Neurological examination confirmed jaw-closing OMD associated with temporomandibular joint osteoarthritis. Previous trials with psychiatric and anticonvulsant medications yielded no benefit. The patient received repeated botulinum toxin injections (100-200U) into the masseter, temporalis, and medial pterygoid muscles, combined with intra-articular platelet-rich plasma (PRP) injections into the temporomandibular joints. This combined strategy led to significant improvement in mouth opening (from 0.5 cm to 3 cm), reduction of joint pain, restoration of prosthesis use, and complete resolution of spasms at one-year follow-up.

Discussion: This case highlights the diagnostic challenges of OMD, frequently misattributed to psychiatric or dental disorders. Botulinum toxin remains the treatment of choice for focal dystonias, providing sustained symptomatic relief. PRP injections may offer additional benefits in patients with concomitant temporomandibular joint pathology, owing to their regenerative and anti-inflammatory properties.

Conclusion: Early recognition of OMD is crucial to avoid misdiagnosis and inappropriate treatment. Combined therapy with botulinum toxin and PRP may represent a promising approach, especially in patients with associated temporomandibular joint osteoarthritis. Further studies are warranted to confirm these findings.

Keywords: *Oromandibular Dystonia; Botulinum Toxin; Platelet-Rich Plasma; Temporomandibular Joint Osteoarthritis; Case Report*

Introduction

Oromandibular dystonia (OMD) is a rare movement disorder characterized by involuntary and sustained contractions of facial, masticatory and lingual muscles, causing involuntary jaw opening or closing. It is a chronic condition that affects speech, swallowing, and eating, causing social embarrassment and negatively impacting daily functioning, mood, and quality of life [1].

The mean time from onset of symptoms to diagnosis in the most common forms of dystonia is 6 years, and this delay is believed to be even longer in oromandibular dystonia as it is often recognized as psychogenic [2].

Case Report

B.T, 52-year-old female patient with a history of depressive disorder under treatment, visited the Department of maxillo-facial surgery with a chief complaint of sustained and prolonged involuntary muscle contractions in the lower and upper jaw, tongue, and face for the past two years. She reported a constant sensation of involuntary teeth clenching and difficulty fully opening her mouth, which significantly affects her ability to eat. As a result, she has to mix her food and consume it through a straw.

Additionally, she struggles with proper jaw and tongue movement during speech, leading to speech difficulties. It is important to note that the patient underwent several dental extractions during childhood due to bruxism and is currently undergoing the process of fitting a dental prosthesis, which was initially placed four years ago (Figure 1).

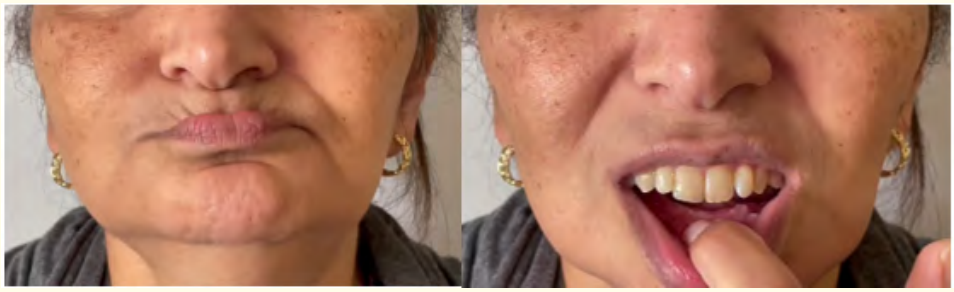

Figure 1: *Jow closing type of OMD, patient uses her finger as sensory trick to allerate dystonia and speak.*

The examination reveals a partially edentulous patient in the upper arch with the presence of teeth 11, 12, 13, 14, 21, 22, 23, 24, 25, and 26, and a completely edentulous lower arch with denture but unable to wear it due to her trismus limiting jaw opening to 0.5 cm and a jaw-closing oromandibular dystonia.

The mouth opening was painful for the patient and palpation of the temporomandibular joints was slightly painful.

The patient did not present with associated blepharospasm and no involvement of cranial nerves or other neurological abnormalities were detected.

The scan shows a decrease in the joint space with preservation of the sphericity of the condylar head on the right and irregularity with osteolysis of the condylar head, without signs of dislocation. These findings are likely indicative of temporomandibular joint osteoarthritis secondary to dystonia (Figure 2).

The patient shared that she had made multiple treatment attempts with various specialists, including an ENT (ear, nose and throat) specialist and a psychiatrist. She was diagnosed as apsychiatric patient, and was prescribed Escitalopram, Benzodiazepine, Clomipramine,

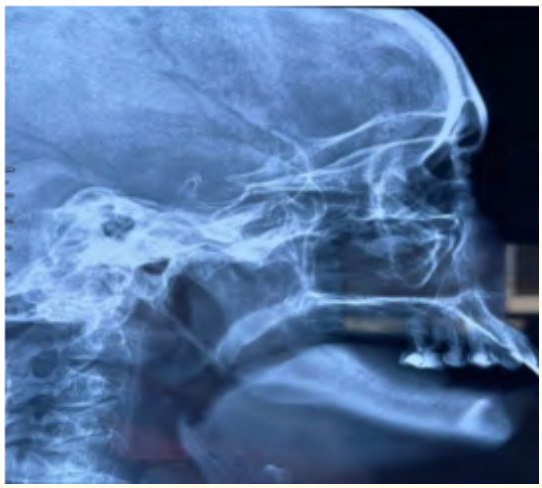

Figure 2: *Ankylosis of the temporomandibular joint.*

Carbamazepine, and Valpoate without improvement for almost 2 years. We treated the patient with botulinum toxin (BTX) injections (100-200 U) in the temporal, masseter, and medial pterygoid muscles every three months. The treatment was reinforced with 2 injections of 2mL of PRP (platelet-rich plasma) at 1-month intervals into the temporomandibular joint to improve mouth opening and alleviate pain. 15-day after the first injection of BTX, there was a notable reduction in spasm frequency, and the mouth opening increased to 1 cm (Figure 3). After the second injection, Joint pain improved and Mouth Opening further increased. At 1 year, the effectiveness of the treatment was confirmed by the absence of spasm and joint pain, the patient was able to wear her lower prosthesis and the month opening increased to 3 cm (Figure 4).

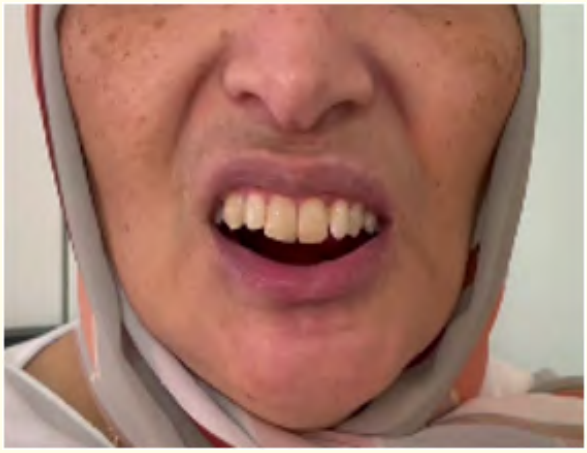

Figure 3: *15 days after BTX injection shows a 1 cm mouth opening with significant decrease of dystonic.*

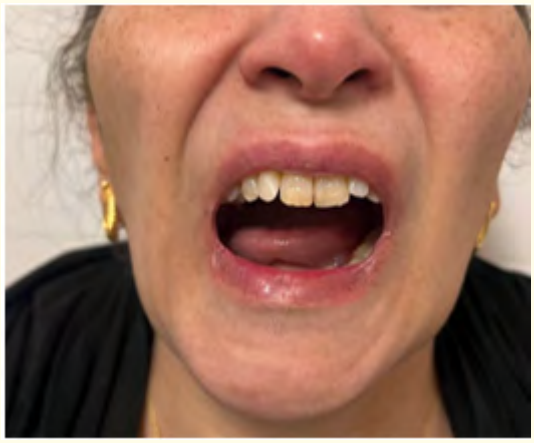

Figure 4: *At 12 mouth follow up: improvement of mouth opening at 3 cm.*

Discussion

Dystonia is considered a network disorder that involves multiple brain regions including basal ganglia, cerebellum, thalamus, and other regions [3]. It can be anatomically categorized according to the affected area as focal, segmental, multifocal, or generalized. Moreover, according to etiology, it can be classified as primary or secondary.

Oromandibular dystonia (OMD) is a rare focal dystonia that mainly affects the tongue, jaw, and mouth. OMD can be clinically divided into jaw opening (JO), jaw closing (JC), mixed (two or more combinations of JO, JC, or jaw deviation), lingual and oro-bucco-lingual subtypes. Although OMD is a focal dystonia, it may also be a part of segmental or generalized dystonia [4,5].

The annual incidence is reported to be between 3.3 and 6.9 per million people [6]. It often occurs after the fifth decade of life and is more common in women.

Idiopathic focal OMD is rare, representing 3 - 5% of all dystonias. Estimated incidence is 3.3/1,000,000 per year and estimated prevalence is 68.9/1,000,000.

OMD is often unrecognized, leading to a delay in diagnosis and treatment [7]. The average time from onset of symptoms to diagnosis in the most common forms of dystonia is 6 years, and this delay is believed to be even longer in OMD [8].

Although isolated OMD is rare, its association with other dystonias is relatively common [9].

Early symptoms of disease are varied and may be subtle, which is believed to contribute to delayed diagnosis and may result in a higher actual prevalence than previously reported [10].

The pathophysiology of dystonia is unclear but is thought to originate in several changes in neuronal activity in striatal circuits such as an alteration in the rate pattern, somatosensory responsiveness, and synchronization of neural activity in palladium thalamocortical circuits [11].

The diagnosis of dystonia is challenging, and is often misdiagnosed as temporomandibular joint syndrome, bruxism, or psychological disturbance. There is no gold standard diagnostic test or biomarker for testing the validity of the diagnosis. It is easier when it is associated with blepharospasm, in the context of Meige syndrome, or other focal dystonias, such as spasmodic torticollis or spasmodic dysphonia.

In our case the classic presentation of the patient in the form of spasmodic contractions with repetitive pattern triggered by occlusion of tooth indicated the jaw closing type of OMD. The exclusive involvement of masseter and temporalis indicated a focal type of presentation [12].

Dental procedures can be considered as a potential source of peripheral iatrogenic injury. Several cases of oromandibular dystonia (OMD) have been reported following dental treatments. Thompson., *et al.* described a case where a woman developed OMD after a dental extraction, Sankhla., *et al.* [13] reported 27 peripherally induced OMD, four of which were wearing new sets of dentures, including one patient with an ill-fitting dental bridge. Among the patients with ill-fitting dentures a habit of manipulation of the jaw muscles to stabilize the new dentures was observed. Hamzei., *et al*. [14] reported the case of a woman who developed facial dystonia within a few hours and severe life threatening laryngeal dystonia with respiratory failure within 3 days after insertion of ill-fitting dentures [15].

Traumatic events in the mouth, such as ill-fitting dentures or multiple extractions, may interfere with the proprioception of the oral cavity, which could contribute to the onset of dystonia. The condition of the muscles progressively worsened over the year. However, it is important to note that the exact causal relationship between these procedures and the development of dystonia remains unclear.

Our patient had indeed undergone several dental extractions and received a prosthesis, both of which may be considered potential trigger factors for her dystonia.

OMD is challenging to manage, and treatment has primarily focused on alleviating the symptoms of the condition. Treatment approaches used to manage OMD include, oral anti dystonic therapies, BTX, local anesthetic blocks, dental appliances, behavioral modification and psychological support.

Oral medication is the usual first line of treatment such as tetrabenazine, diazepam, and carbamazepine. Anticholinergic drugs reduce muscle spasm by centrally inhibiting the parasympathetic system. Benzodiazepine decreases monosynaptic and polysynaptic reflexes by increasing presynaptic GABA inhibition, a similar action to Baclofen. Anticonvulsants such as carbamazepine reduce severe muscle spasm by decreasing polysynaptic response [16].

After medications, the other primary method for treating dystonia is chemodenervation using botulinum toxin. Botulinum toxins exert their therapeutic benefit by blocking the release of acetylcholine into the neuromuscular junction [17]. In 1989, Blitzer and colleagues [18] first described the injection of botulinum toxin for oromandibular dystonia. They described injecting many of the orofacial muscles in oromandibular dystonia and claimed that masseter, temporalis and medial pterygoid muscle injections helped with suppressing the overall oromandibular dystonia. Effects generally take effect in the first 2 weeks and last for 3 - 4 months.

Since its introduction in the 1980s, botulinum toxin has revolutionized the treatment of dystonia. Botulinum toxin is a toxic protein produced by the bacterium *Clostridium botulinum*, which exists in seven different serotypes (toxins A - G) [19]. The toxin is injected into dystonic muscles, thereby weakening the muscles and ameliorating dystonic symptoms.

Our patient, BTX improve mouth opening and their ability to eat and speak.

Although a number of alternative treatments have been proposed for OMD. Neurosurgical procedures including deep brain stimulation [20], muscle afferent block therapy, and acupuncture [21]. There are insufficient studies comparing these procedures head-to-head. Further research on treatment strategies is warranted.

Platelets secrete numerous growth factors, including insulin-like growth factor (IGF), transforming growth factor beta 1 (TGF-β1), platelet-derived growth factor (PDGF), as well as cytokines. These regulate inflammation, proliferation, and tissue remodeling during healing and regeneration [22].

At the cellular level, *in vitro* studies have shown that after the administration of PRP (platelet-rich plasma), there is a proliferation of chondrocytes, fibroblasts, osteoblasts, and mesenchymal stem cells, leading to collagen deposition in cartilage repair and bone remodeling. Furthermore, PRP stimulates synoviocytes, increasing the secretion of hyaluronic acid, which is important for joint lubrication [23].

For about a decade, autologous blood derivatives such as PRP (platelet-rich plasma) and PRF (platelet-rich fibrin) have been widely studied and used in regenerative medicine to treat tendinopathies, assist in the healing of bone grafts and in plastic surgeries.

In the field of dental surgery, PRP is already used to promote tissue healing in maxillofacial reconstructions, sinus augmentations, periodontal grafts, and implant surgeries [24].

Intra-articular injection of PRP in the treatment of temporomandibular joint (TMJ) osteoarthritis significantly reduces pain compared to hyaluronic acid, placebo, or no injection, and may increase mouth opening capacity. Several studies recommend an injection of 2 mL of highly concentrated and highly purified PRP [25].

Moreover, our patient also had TMJ osteoarthritis, which benefited from intra-articular PRP injections. This procedure was further supported by a better understanding of the hemostatic and regenerative roles of platelets.

PRP, acting as a broad vector of growth factors, should find its place among intra-articular treatments for osteoarthritis. However, it is not yet mentioned in the main recommendations of various rheumatology organizations [26].

In our patient, two sessions were performed at two-week intervals, with a total of 2 mL of PRP injected at each session (1 mL per joint). This strategy, inspired by the protocols proposed by Guarda-Nardini., *et al*. (2012) and Limchaichana., *et al*. (2022) [27,28] aimed to modulate joint inflammation, enhance the trophic support of periarticular tissues, and optimize mandibular biomechanics in the context of neuromuscular dysfunction. The patient reported a significant reduction in joint pain, improved mouth opening range, and better mandibular coordination. The PRP/botulinum toxin combination thus appears promising for mixed presentations involving both muscular and articular components.

Conclusion

OMD is particularly disabling because it interferes with the ability to eat and speak, and may be associated with marked discomfort. Because of its rare occurrence, patients with OMD are probably often misdiagnosed, or recognized as psychogenic and there are delays in its diagnosis and treatment. Consequently, these patients may also receive incorrect dental treatment, and the symptoms may worsen over the years. Botulinum toxin has been proven to be superior to medical treatment particularly in focal dystonias. The use of PRP could slow down the arthritic process in addition to pain management.

Bibliography

1. Jinnah HA and Factor SA. "Diagnosis and treatment of dystonia". *Neurologic Clinics* 33.1 (2015): 77-100.
2. Tan EK and Jankovic J. "Epidemiology and clinical spectrum of primary dystonia". *Journal of the Neurological Sciences* 163.2 (1999): 219-230.
3. Neychev VK., *et al*. "The functional anatomy of dystonia". *Neurobiology of Disease* 42.2 (2011): 185-201.
4. Bhidayasiri R and Marsden CD. "Oromandibular dystonia: a clinical study of 115 cases". *Movement Disorders* 9.2 (1994): 119-126.
5. Jankovic J. "Treatment of dystonia". *Lancet Neurology* 5.10 (2006): 864-872.
6. Tan EK and Jankovic J. "Phenomenology of oromandibular dystonia: a study of 100 patients". *Archives of Neurology* 56.3 (1999): 290 296.
7. Yoshida K. "Clinical features of oromandibular dystonia and their treatment". *Movement Disorders* 21.12 (2006): 1893-1897.
8. Jinnah HA and Factor SA. "Diagnosis and treatment of focal dystonias". *Lancet Neurology* 14.3 (2015): 267-278.
9. Albanese A., *et al*. "Phenomenology and classification of dystonia: a consensus update". *Movement Disorders* 28.7 (2013): 863-873.
10. Defazio G., *et al*. "Clinical characteristics of patients with isolated or combined dystonia". *Movement Disorders* 28.10 (2013): 1478-1485.
11. Berardelli A., *et al*. "Pathophysiology of dystonia: a review of evidence". *Brain* 121.7 (1998): 1195-1212.
12. Tan EK and Jankovic J. "Clinical approach to oromandibular dystonia". *Current Opinion in Neurology* 12.4 (1999): 385-390.
13. Sankhla C., *et al*. "Peripherally induced dystonia". *Neurology* 51.3 (1998): 768-774.
14. Hamzei F., *et al*. "Acute onset oromandibular dystonia following dental procedures". *Movement Disorders* 16.5 (2001): 985-987.
15. Hamzei F and Jabbari B. "Severe laryngeal dystonia induced by dental prosthesis". *Neurology* 58.1 (2002): 147-149.
16. Jankovic J and Tintner R. "Pharmacological treatment of dystonia". *Lancet Neurology* 358.9283 (2001): 917-924.
17. Simpson DM., *et al*. "Assessment: Botulinum neurotoxin for the treatment of movement disorders". *Neurology* 86 (2016): 1818-1825.
18. Blitzer A., *et al*. "Botulinum toxin injection for oromandibular dystonia". *Annals of Otology, Rhinology and Laryngology* 98.9 (1989): 705-710.
19. Rossetto O and Montecucco C. "Mechanism of action of botulinum neurotoxins". *Journal of Neurochemistry* 104.2 (2008): 287-301.
20. Vesper J., *et al*. "Deep brain stimulation in dystonia: a review". *Neurosurgical Review* 32.4 (2009): 415-426.
21. Sethi KD. "Surgical treatment of dystonia: a review". *Neurology India* 49.4 (2001): 329-336.
22. Marx RE. "Platelet-rich plasma: evidence to support its use". *Journal of Oral and Maxillofacial Surgery* 62.4 (2004): 489-496.
23. Anitua E., *et al*. "Platelet-rich plasma: a review of its biologic features and clinical applications". *Journal of Tissue Engineering and Regenerative Medicine* 1.4 (2006): 245-259.

24. Marx RE. "Platelet-rich plasma: growth factor enhancement for bone grafts". *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology* 97.6 (2004): 756-763.

25. Xie X., *et al*. "Efficacy of platelet-rich plasma in temporomandibular joint osteoarthritis: a systematic review". *Journal of Cranio-Maxillofacial Surgery* 47.11 (2019): 1746-1753.

26. Filardo G., *et al*. "Platelet-rich plasma intra-articular injections for the treatment of osteoarthritis: literature review and evidence-based recommendations". *Blood Transfusion* 16.3 (2018): 217-226.

27. Guarda-Nardini L., *et al*. "Platelet-rich plasma injection for TMJ disorders". *Journal of Oral and Maxillofacial Surgery* 70.12 (2012): 2835-2841.

28. Limchaichana R and Chuenkongkaew T. "Platelet-rich plasma in temporomandibular joint dysfunction: clinical outcomes". *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology* 133.2 (2022): e91-e98.

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