

Review of Literature**Etiology, Diagnosis and Management of Oromandibular Dystonia:
an Update for Stomatologists**Saeed Raofi¹, Hooman Khorshidi¹, Maryam Najafi¹¹ Dept. of Periodontology, School of Dentistry, Shiraz University of Medical Sciences, Shiraz, Iran.**KEY WORDS**

Botulinum Toxin;
 Diagnosis;
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ABSTRACT

Oromandibular dystonia (OMD) is a rare focal neurological disorder that affects mouth, face, and jaws. This comprehensive literature review aimed to summarize the current evidence for etiology, diagnosis, and management of OMD and assess the possibility of dental origin of the disease and dental treatment plans for these patients.

Different online databases namely PubMed, Google scholar, and Scopus were searched. The keywords “oromandibular dystonia”, “orofaciomandibular dystonia”, “orofacial-buccal dystonia”, “lingual dystonia”, “jaw dystonia”, “cranial dystonia”, and “adult-onset facial dystonia” were searched in the title and abstract of publications from 1970 to 2016. The inclusion criterion was the dental etiology and/or dental treatment. Out of 1260 articles, only 37 articles met the inclusion criteria. OMD can be caused or exacerbated through different dental treatments within which anyone is likely to be involved due to various reasons. Some novel methods employed to relieve this syndrome have led to certain cure or improvement of symptoms in several cases. OMD patients may refer to dentists with involuntary jaw movements and intraoral presentations. Thus, the dentists should be aware of the symptoms and signs and refer the suspicious cases. Dentists should also be familiar with special considerations when managing OMD patients.

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Introduction

Dystonia is the manifestation of involuntary lasting severe muscle contractions, which lead to rhythmic and atypical movements in different parts of the body. 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 etiology. Primary dystonia is idiopathic or inherited; while, secondary dystonia advances after traumatic or surgical incidents, brain diseases, and medications. [1]

That type of dystonia that involves the oral cavity

is described as oromandibular dystonia (OMD). It is a rare focal neurological disorder that affects the lower facial muscles. It is characterized by repetitive or sustained involuntary prolonged spastic movements of the tongue, facial, and masticator muscles. OMD is classified as jaw opening, jaw closing, jaw deviating, or lingual dystonia, or a combination of these. [2-4] Combination of OMD, belpharospasm, and dystonic movements of the upper face is called Meige's syndrome. [5-6] Tongue protrusion is usually a component of OMD or Meige's syndrome. [7] Other terms for OMD are orofaciomandibular dystonia, orofacial buc-

cal dystonia, lingual dystonia, jaw dystonia, cranial dystonia, and adult onset facial dystonia. [2-5]

The prevalence of OMD has been reported to be as high as 6.9/100,000 cases and the incidence has been reported up to 3.3 cases per million. [4] It has been suspected that dental and oral surgery interventions or treatments can be associated with the onset of dystonia and ethical considerations in dentistry [8] prompted us to review related papers.

The purpose of this study was to review the literature to assess the possibility of dental origin of the disease and dental treatment plans for these patients.

Search Strategy

Different online databases such as PubMed, Google Scholar, and Scopus were investigated in English. The randomized controlled trials, case-control studies, case series, and single case reports containing a report on a clinical outcome were included. In addition, the reference list of these articles was searched and those considered important were selected, as well.

A comprehensive literature review was done from 1970 up to December 2016. The searched terms were "oromandibular dystonia", "orofaciomandibular dystonia", "orofacial- buccal dystonia", "lingual dystonia", "jaw dystonia", "cranial dystonia", and "adult-onset facial dystonia", per se and in combination with "dental treatment" and "dental origin". The inclusion criterion was the dental etiology and/or dental treatment. Out of 1200 articles, only 37 articles met the inclusion criteria. Information collected from these 37 references was used to write the literature review.

Discussion

Clinical manifestations

Medical signs and symptoms

The onset of symptoms is usually between the ages of 40 to 70 years and is more common in women. The symptoms only occur during activities such as speaking or mastication. Patients usually report triggers like stress, talking, chewing something and praying. [9] Usually, routine laboratory tests are normal. [4] In most cases, the oral function is reported to be compromised and associated with social embarrassment, reduced quality of life, depression, and weight loss.

Severity of symptoms and progression of the disease are more prominent in post-traumatic OMD

than in primary OMD; however in the former, there is lower tendency to spread to contiguous or non-contiguous segments. Family history of movement disorders is also less frequent in post-traumatic OMD than in primary OMD. Specific activities or sensory stimuli, sometimes called sensory tricks are used to relieve the dystonia in OMD, but not in other type of dystonia, such as cervical dystonia. [5]

Dental and facial signs and symptoms

Clinical presentations depend on the affected muscles, as well as the severity and distribution of OMD. [3] Dysfunctions include impaired mastication, dysphagia, speech alterations (dysphonia), unconscious opening and closing of the mandible, pulling and twisting of the mandible forward or laterally, and temporomandibular disorders (TMD) such as open locks. [4, 10]

Dystonic spasms may be seen as nasal contractions, facial grimacing, lip pursing or sucking, bruxism, tongue dyskinesia, mouth corner retractions, and platysma spasms. Dysarthria or breathing difficulties are also reported rarely. [2-3, 11]

The difference between idiopathic sleep bruxism and OMD-induced bruxism is that the latter usually stops while sleeping. [4] Watts *et al.* [12] assessed the plausible relationship between bruxism and cranial-cervical dystonia. The prevalence of bruxism was higher in patients with cranial-cervical dystonia. Nevertheless, this difference was not significant between other diseased groups and the controls. Trismus, bruxism, and forceful involuntary jaw closure or temporomandibular joint (TMJ) dislocation can lead to trauma and damage of the oral cavity structures, dental restorations, and dentures, excessive dental wear, dental fractures, and trauma of the lips, gums, and tongue; while, jaw-opening dystonia may be associated with TMJ overload. [4, 7, 9, 11]

Muscle pain is usually associated with headache and facial pain as a result of dystonic activity and forceful contractions. However, most patients usually complain of muscular tension or tiredness, and pain was quite rarely reported. In a review by Blanchet *et al.*, [13] pain was reported in approximately 20% of edentulous patients, indicating it to be probably related to structures, not the muscles. The dystonic activities are also associated with tooth loss and hyposalivation.

Etiology

The mechanism and the cause of OMD are not well known. OMD may occur per se as a neurological disorder (with or without a hereditary history), or as secondary to certain drugs or disorders such as trauma or Wilson's disease. [2, 7] Central nervous system (CNS) trauma, neuroleptic exposure, hypoxic brain damage, metabolic disorders, and ischemic or demyelinating lesions in the upper brain stem can be related to OMD. [5]

Michelotti *et al.* [14] reported a 30-year-old woman affected with OMD with a 12-year follow-up. They reported temporal pattern of symptoms in patient and discussed the possible relationship between OMD and hormonal factors. Peripheral trauma is a known cause or a predisposing factor in different neurological and movement disorders like Parkinson's disease, tremors, dystonia, painful legs and moving toes; this relationship has been documented in many reports. [5] The time between the initiation of trauma and the onset of OMD is variable. Peripherally-induced dystonia is usually not recognized, especially if the trauma is relatively trivial or the duration between the trauma and the onset of dystonia lasts longer than a few days. [5, 15]

Fletcher *et al.* [16] reported that peripheral injuries may affect basal ganglia function and provoke the onset of dystonic movements in carriers of ITD (idiopathic torsion dystonia) gene (genetically predisposed individuals). Despite the comprehensive document on peripherally-induced dystonia caused by injury to the affected body part, OMD from orofacial or dental procedures is scarcely reported in dental literature and neurological articles. [15] Thus, its precise prevalence is unknown. Although some cases of OMD have been reported after dental procedures, the causal relationship between these procedures and dystonia is still unclear. [3]

Sutcher *et al.* [17] reported four patients who wore ill-fitting full dentures that resulted to jaw-opening OMD. The patients had worn the dentures from a minimum of 1 to many years before noticing the abnormal movements. Sankhla *et al.* [18] reviewed 9083 patients with movement disorders, 197 of which were diagnosed with OMD. In 27 cases, the onset of the symptoms of OMD was related to a prior trauma of the face or mouth. Ten cases had family history of movement disorders, prior exposure to neuroleptic drugs, and

associated dystonia affecting other regions. Out of the 27 cases, 21 had prior dental procedures (new ill-fitting full or partial dentures, root-canal therapy, gingivectomy, crowns, tooth removal, apicoectomy, osteotomy, or TMJ arthroscopic surgery). They concluded that different predisposing factors such as an associated movement disorder, family history of tremors, edentulous state, exposure to neuroleptic drugs, and peripheral nerve injury may contribute to the development of this movement disorder under some conditions or in certain vulnerable people; however, the relationship between them may be purely coincidental.

Thompson *et al.* [19] reported a female individual who developed jaw dystonia following dental extraction. Jankovic and Van der Linden [20] assessed dystonia and tremor induced by peripheral trauma and predisposing factors. They noticed that in 65% of cases, possible predisposing factors might have caused the pathogenesis of the trauma-induced abnormal involuntary movements. Peñarrocha *et al.* [21] reported a case of OMD with belpharospasm, tooth loss, and occlusal alterations that exacerbated the dystonic movements. They believed that tooth loss might trigger or worsen the dystonia.

Hamzei *et al.* [22] reported a woman with a history of Parkinson, who developed jaw-closing dystonia within 8 hours of ill-fitting full denture insertion. The dystonia spread very fast to involve the larynx, causing life-threatening laryngospasm which was treated by intubation. They concluded that patients with craniocervical dyskinesia might be at risk of developing severe and rapidly-spreading dystonia as a result of dental procedures. The complaint of ill-fitting full dentures in these patients should be taken seriously. Thorburn and Lee [15] discussed two OMD cases which occurred following extractions and full dentures. Balasubramaniam *et al.* [23] discussed a case of OMD following dry socket associated with third molar surgical extraction. Seeman *et al.* [24] reported a case of transient OMD following a dental filling in a woman receiving quetiapine (a second-generation antipsychotic). They suggested that the dental procedure might have caused a triggering effect.

Chidiac [10] described a case of OMD where an open lock of the mandible with a constant protrusive posture was persisting for more than 3 months. Ac-

ording to the patient's history, the problem began during a dental treatment for adjusting the occlusion. In the reported case, spasm of lateral pterygoid muscles was revealed by neurologic evaluation, and medication and botulinum neurotoxins (BoNT) were utilized for relief. The author noted a huge loss of vertical dimension, which was restored through fabricating a provisional denture.

Bakke *et al.* [4] reported 21 cases of OMD, 2 of which were related to injury or infection. No detail was mentioned about the kind of trauma and infection. Chung *et al.* [25] published a case report of dental implants-induced task-specific OMD in 2013. The patient presented a 1-year history of involuntary retraction of lips while speaking, as well as dental implant surgery 6 months earlier. There was neither medical history or medications, nor excessive use of perioral muscles due to the patient's job or hobby. It was concluded that there might be a potential causal relationship between dental implants as peripheral injury and the development of OMD.

Diagnosis

The diagnosis of OMD is clinical and complicated, since it presents in various forms and severities. It responds to no diagnostic medical test; hence, the diagnosis is based on individual information, history, neurological examination and confirmation by intramuscular electromyography (EMG). OMD patients are usually diagnosed by neurologists and are aware of their problems. [18] The differential diagnosis includes TMJ disorders (like bruxism or spontaneous condylar dislocation), hemifacial spasm, and psychological disorders. The symptoms of OMD can get worse by emotional factors, which is among the reasons for the delayed diagnosis.

Treatment

Treatment of OMD is multi-disciplinary and varies from one patient to another.

Medication

There is inadequate evidence-based information about the efficacy of various medications currently being used for dystonia. [5] Nevertheless, anticholinergic, baclofen, benzodiazepines, antiparkinson drugs, anti-convulsants, carbamazepine, dopamine receptor antagonists, levodopa and lithium are some of the agents used to manage OMD. [26]

Physiotherapy

Physiotherapy is thought to promote brain re-wiring over time, so as to reduce the dystonic movements. This response is well recognized in musicians. [15]

Botulinum neurotoxins (BoNT) injections

BoNT injection, a promising therapy for treating OMD, is a potent neurotoxin that blocks the release of acetylcholine at the presynaptic junction, resulting in temporary chemical denervation of skeletal muscles. BoNT injections can leave even better effects if guided with EMG, which allows longer intervals between the BoNT injections. BoNT injection is mainly technique-dependent. Depending on the dose of therapy, jaw weakness or tremor, loss of smile, and dysphagia are expected as the side effects. BoNT may be immunogenic, and some patients may develop secondary non-responsiveness following multiple injections. [27] Swallowing difficulties, speech problems, and excessive muscle weakness have been reported in the treatment of lingual dystonia by BoNT. [11, 21]

Muscle afferent block (MAB)

Yoshida *et al.* [28] found MAB to be highly effective on OMD; although, it had no effect on patients with dyskinesia symptoms. Thus, OMD and oral/orofacial dyskinesia might differ in physiopathology. MAB is a promising means of treating OMD, which is less expensive and cause no major side effect or resistance to the therapy.

Operative therapies

There is no specific surgical therapy for OMD, and the role of CNS procedures has not been proven. [15] In some patients with peripherally-induced OMD, arthroscopy and other TMJ surgeries were done with no useful outcome; the conditions got even worse in some cases. [21] Yoshida [29] performed surgical resection of the bilateral coronoid processes in 2 patients with jaw-closing dystonia and severe trismus. Treatment with medication, BoNT injection, and MAB therapy were not effective, and no significant enlargement of the bilateral coronoid process was observed. The jaw opening increased to 50 mm in both patients. The final coronoidotomy might be a useful treatment for patients with quite severe jaw-closing dystonia, where other conservative therapies are ineffective.

Other therapeutic approaches

Psychosocial and occupational therapy, support groups

participation, cognitive behavioral therapy, [15] and deep brain stimulation surgery were also designed to reduce the hyperactivity of the muscles. [1] Deep brain stimulation surgery was reported as an effective and safe treatment. Pallidotomy, thalamotomy, and focused ultrasound lesioning are the other options for patients with dystonia. [30-31] Dressler *et al.* [32] reported that frontalis suspension operation was a nonthreatening procedure producing strong and constant satisfying effects on eyelid opening apraxia in belparospasm. According to Xu *et al.* [33] and Peeraully *et al.*, [34] acupuncture was reported to be helpful. Although it did not work as rapidly as the local injection of Botox, their efficacy might be similar. Moreover, this treatment had no adversarial reaction.

Dental treatments and OMD

Searching the databases yielded a few articles on OMD patients who received dental management. Watt *et al.* [1] published a case report on the treatment of OMD in a patient who presented a sensory trick to reduce his symptoms. They made a suitable removable dental appliance to mimic the sensory trick. Its positive effects might be due to the proprioceptive stimulation. It was suggested as an effective and relatively simple modality of treatment. Gonzalez-Alegre *et al.* [9] in a retrospective chart review of 27 OMD patients reported that a sizable proportion of them exhibited sensory tricks that could be utilized to develop therapeutic prosthetic appliances.

Thorburn and Lee [15] found intraoral sensory tricks in two cases of OMD following extractions and full dentures; therefore, acrylic shapes were made to mimic the tricks. Arthroscopy and other TMJ surgeries yielded no useful result in some patients with peripherally-induced OMD, and even exacerbated the conditions in some others. [21] Yoshida [29] performed surgical resection of the bilateral coronoid processes for two patients with jaw-closing dystonia and severe trismus. Then, bilateral coronoidotomy and masseter muscle stripping was done in 18 cases. A mean overall improvement was detected in patients' symptoms. [35]

Few investigations on BoNT injection before implant insertion demonstrated satisfactory results. In particular, Sibley [36] reported a case of edentulous OMD restored with implants. The author suggested that the conventional techniques such as delayed loading,

axial implant placement and BoNT injections could be used to reduce the dystonic movements with more predictable rehabilitation plan.

Schneider and Hoffman [11] utilized prosthesis with minimal adjustments in recalls for a case of OMD. The patient was pleased and able to maintain a comfortable facial position and her speech was intelligible. Complete resolution of symptoms while wearing the prosthesis was observed. The TMJ remained asymptomatic during this period. At the 3-month recall, the symptoms had returned with less intensity. By modification of the prosthesis, slight improvement of the symptoms was achieved. Blanchet *et al.* [13] adopted a flat-plane occlusal splint in some cases to reduce the tissue trapping during spasm. They announced the plausible existence of causal relationships between dental procedures and OMD, since dental treatment can alter the sensory input.

Peñarrocha *et al.* [21] reported that mandibular overdentures supported by endosteal implants were satisfactory in reestablishing the occlusion, prosthetic stability and improvement in the masticatory muscles movements. During the 5-year follow-up, stabilization of the oromandibular dystonic movements, and improvement of function and esthetic were observed; but, the belparospasm got worse. It was not reported whether the implant or other interventions of maxillary sinuses triggered dystonia. [37] Jang *et al.* [38] reported two cases of OMD after dental extractions, with no family history of movement disorders, organic brain lesion, or exposure to neuroleptic drugs. Immediate implantations of the lower right molar teeth in one case, and partial denture in the other were performed. They suggested that the onset of dystonia might have been caused by the dental intervention.

It was reported that paroxysmal focal dystonia usually arises alongside the other manifestations of neuro-Behçet's disease; [39] however, the present review could not find any article on the relation between the oral health status and oromandibular dystonia. [40]

Brissaud *et al.* [41] reported a repeated tongue biting in a 17-month-old girl. The dental managing included tongue suturing, tongue protection with bite guard, composite restorations for shaping sharp teeth, partial glossectomy, with no dental extraction. The bite guard was adjusted to accommodate the newly erupting

Table 1: The articles categorized based on dental etiology

Article	The number of studied cases	Dental etiology that trigger or worsen the dystonic movements (based on patients' reports)	Duration of OMD or dystonia
Jang, 2012	2	Dental extraction	-
Penarrocha, 2001	1	Loss of teeth and occlusal alteration	2 years
Hamzei, 2003	1	Ill-fitting denture	Facial dystonia: 3 hours; Laryngeal dystonia: 3 days
Thorburn, 2009	2	Dental extraction and full denture	1 case: 3weeks The other: 6 months
Balasubramaniam, 2008	1	Oral surgery	-
Seeman, 2008	1	Dental filling	8 weeks
Chidiac, 2011	1	Occlusal adjustment	
Chung, 2013	1	Dental implant	1 year
Sutcher, 1971	4	Ill-fitting denture	1 year to many years
Sankhla, 1998	21	Ill-fitting new denture, root-canal therapy, gingivectomy, crowns, tooth removal, apicoectomy, osteotomy, TMJ arthroscopic surgery	Range: 1-16 years
Thompson, 1986	1	Dental extraction	-
Yoshida, 2006	2	One case: occlusal splint The other: psychogenic disease	1 case: 8 years The other: 4 years

teeth. Botox infiltrations and antidystonic medication were administered for controlling the dystonia.

Zuza *et al.* [42] reported a case with generalized idiopathic dystonia who received periodontal treatment.

Motor limitations caused difficulty in plaque control and subgingival caries. They adopted the methods used for oral hygiene by using Electrical toothbrush and dental floss fork. The good results allowed for an increase in the dental crown in teeth with subgingival caries.

Despite the limited articles, a comprehensive overview was achieved on the dental origin and dental treatment of this syndrome (Table 1 and 2). The clinicians should consider the possibility of OMD in cases of parafunctional movements such as bruxism and clenching. The OMD patients may present to dentists with involuntary jaw movements and intraoral findings/problems such as inability to bite in intercusp occlusion, attrition of the teeth, and trismus. Patients with OMD may also suffer from hyposalivation; nevertheless, the pooling of saliva on intraoral examination was reported. [6] Thus, the dentists should be aware of the special consideration in managing dental treatment of OMD patients; and frequent dental visits seem desirable to support the masticatory function in prosthetic teeth replacements.

Patients may not be aware of their problem and it can be misdiagnosed as a dental problem like denture problems, bruxism, or TMD; [2, 11] therefore, the dentist should be familiar with signs and symptoms of OMD and refer the patients for more assessments.

Sometimes the patients have found certain sensory tricks which can help them control or suppress the dystonia. Finding intraoral sensory trick and fabrication of prosthetic devices can help dental professionals to treat the symptoms of OMD. Surgical therapies are the last effort for individuals suffering from certain types of dystonia. Based on the available data, it seems that there is no specific surgical therapy for OMD.

Prosthetic treatment planning after controlling the problems of involved muscle with modalities such as BoNT injection yields satisfactory results. BoNT treatments of the involved muscles have led to marked improvements; [43-46] thus, the dentists are required to be familiar with methods of Botox injections. They should recognize the signs and symptoms of OMD when considering the differential diagnosis of TMJ disorders. According to the therapeutic use of BoNT, it is recommended that training should be at least included in the curriculum of some postgraduate specialties in dentistry.

Conclusion

Duo to various dental treatment options, all people are somehow being involved in dental treatments, with which OMD is likely to coincide. Dental treatments can be the cause of onset or exacerbation of OMD. In addition, the novel methods adopted by dentists to relieve this syndrome have resulted in certain cure or improvement of the symptoms in many cases.

Since the OMD patients may present to dentists with involuntary jaw movements and intraoral finding/

Table 2: The articles categorized based on dental treatment

Article	The number of studied cases	Duration of OMD or dystonia	Dental treatment for OMD	Dental treatment outcomes
Peñarrocha, 2001	1	2 years	Dental implant-supported overdentures	Effective in stabilizing the oromandibular dystonic movements
Thorburn, 2009	2	1 case: 3 weeks The other: 6 months	Acrylic shapes	Effective
Watt, 2014	1	6 years	Removable dental appliances	Effective
Gonzalez-Alegre, 2014	27	-	Prosthetic devices	Effective
Yoshida, 2006	2	1 case: 8 years The other: 4 years	Bilateral coronoid resection	Complete treatment of OMD
Sibley, 2013	1		Dental implant	Effective
Schneider, 2011	1	2 years	Prosthesis with minimal adjustment in recalls	Effective
Blanchet, 2005			Flat-plane occlusal splint	Effective
Yoshida, 2016	18	41.1±44.7 months	Bilateral coronoidotomy and masseter muscle stripping	Effective
Zuza, 2016	1		Oral hygiene improving	Effective
Brissaud, 2016	1		Tongue suturing, tongue protection with bite guard, composite restorations for shaping sharp teeth, partial glossectomy, no dental extractions	Effective

problems, the dentists should be aware of the symptoms and signs to refer the suspicious patients if necessary.

They are also required to be familiar with the special considerations in managing dental treatment of OMD patients. To progress the current medical and related health care options for patients with dystonia, excellent trials that study the efficacy of treatments are needed to be done.

Conflicts of Interest

The authors declare no conflict of interest.

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