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Oromandibular Dystonia: An Update for Dental Professional

Shruthi Hegde[1-a]*, Vidya Ajila[2-b]

[1]Department of Oral Medicine and Radiology, AB Shetty Memorial Institute of Dental Sciences (ABSMIDS), Deemed to be University, Mangalore, India.
**Corresponding author*

Reviews

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ABSTRACT

Oromandibular Dystonia (OMD) is a type of focal dystonia which affects the masticatory, cervical, facial, eyelid, laryngeal, and pharyngeal muscles. OMD patients may consult dentists with involuntary movement or spasm of the lips, tongue, involuntary jaw opening or closing, changes in the occlusion, slurred speech, drooling of saliva, difficulty in mastication, swallowing, and speaking. Due to the uncommon occurrence of OMD, this condition may be misdiagnosed and may lead the patients to exposure to unnecessary treatments. Thus, thorough knowledge regarding the features of OMD is essential for the dentists. This review will present the diagnosis and management of OMD, focusing on its signs and symptoms.

Keywords: Oromandibular Dystonia, Botulinum Toxin, Oral Appliances.

[a] *drshruthihegde@yahoo.co.in* *https://orcid.org/0000-0002-0744-5593* [b] *drvidyaajila@nitte.edu.in* *https://orcid.org/0000-0002-5744-9322*

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Introduction

Dystonia is a neurological disorder which is manifested as sustained involuntary muscle contractions leading to abnormal repetitive movements in various body parts.[1,2] Clinical presentation varies depending upon the affected musculature, severity and distribution.[3] The causative factor for this condition may be hereditary, birth-related, reaction to certain drugs, physical trauma, infection or poisoning.[4] According to the affected regions, dystonia can be categorized as focal, segmental, multifocal, and generalized.

Oromandibular Dystonia (OMD) is a type of focal dystonia which affects the masticatory, cervico-facial, eyelid, pharyngeal and laryngeal muscles.[5] OMD can have symptoms of involuntary movement/spasm of the lips, tongue, lip pursing involuntary jaw opening or closing. Muscle hyperactivity and resulting fatigue can cause muscle pain. Other symptoms noticed in orofacial region are dysphagia, dysphonia, deviation of the mandible, subluxation of the temporomandibular joint and intraoral soft-tissue trauma. Change in the occlusal relationship, excessive drooling, problems with mastication, swallowing, and speech may be noticed.[3,5-7] Due to the uncommon occurrence of OMD, this condition may be misdiagnosed and it may lead to unnecessary treatments. OMD patients may consult dental professionals with intraoral presentations and involuntary movements of jaw.

The aim of the present study was to perform a systematic review of the literature over the past 10 years in "PubMed" and "Science Direct" database for studies, case reports and reviews addressing signs and symptoms of OMD, diagnostic criteria and dental management of these patients.

Methods

This systematic review was prepared in accordance with the Preferred Reporting Items for Systematic Reviews and Meta Analyses (PRISMA) statement (Figure 1).[8]

Search Strategy

Literature search was carried out in "PubMed" and "Science Direct" database with search terminology "oromandibular dystonia" and "dental treatment". Data available from 2010 to 2020 were selected. English-language articles were chosen for the study. Review articles, clinical studies, single case reports and case series containing information regarding clinical features, diagnosis and management were included. In addition, the reference list of these articles was searched and those considered important were selected, as well. Only full text available was selected. Our search resulted in a total of 101 articles. In which 49 articles from PubMed database and 52 articles from Science Direct database. In our review 76 articles were excluded after reading the

title, abstract and duplication. Articles submitted to full-text analysis for eligibility were 25 of them. Out of which 21 articles fulfilled eligibility criteria. Studies included in final review for summarizing clinical features and management strategies used were 12 of them.

Results

Out of 12 studies included for final review following data were extracted: author, publication year, number of cases reported, age, gender, clinical features, treatment summary and outcome (Table 1).[4,7,9-17] The data was tabulated and the results recorded. Total of 89 patients with OMD were reported in 12 articles. Total of 44 female patients (49%) and 45 male patients (51%) were reported in the included studies. Age of the affected individuals ranged from 25 to 84 years. Out of 89 patients 82(92%) of them were treated with botulinum toxin A. Out of 8 studies reported use of botulinum toxin A for their patients, 6 studies reported improvement, one study reported complete improvement and in one study outcome was not reported. Other treatment modalities reported were dental prosthesis, occlusal stabilization appliance, occlusal splint, physical therapy and drugs such as baclofen, analgesics, anticholinergics, carbamazepine. All the studies reported improvement of symptoms.

Discussion

This systematic review was performed to evaluate clinical features, diagnostic criteria and dental management of cases reported with OMD from 2010 to 2020. Uncommon encounter of dental professionals with OMD patients in dental office may pose difficulty for diagnosis and treatment plan. Therefore, present review focuses on detailed discussion of the clinical features, etiology, classification, diagnosis, differential diagnosis, treatment and dental consideration of OMD patients.

According to the American Academy of Oral Medicine, OMD is a movement disorder associated with uncontrolled contraction of the affected muscles, leading to abnormal posture and functional difficulties, including psychosocial withdrawal.[18]

OMD is a rare condition. The incidence has been reported as 3.3 cases per million persons and prevalence is around 6.9 per 100,000 people. This systematic review also revealed only 89 reported cases of OMD over past 10 years. The onset of symptoms is usually seen in the age group of 40 to 70 years. In our study reported age range was 25 to 84 years. According to the literature women are commonly affected than men.[9,19] However, this review slight male predominance (51%) was noted.

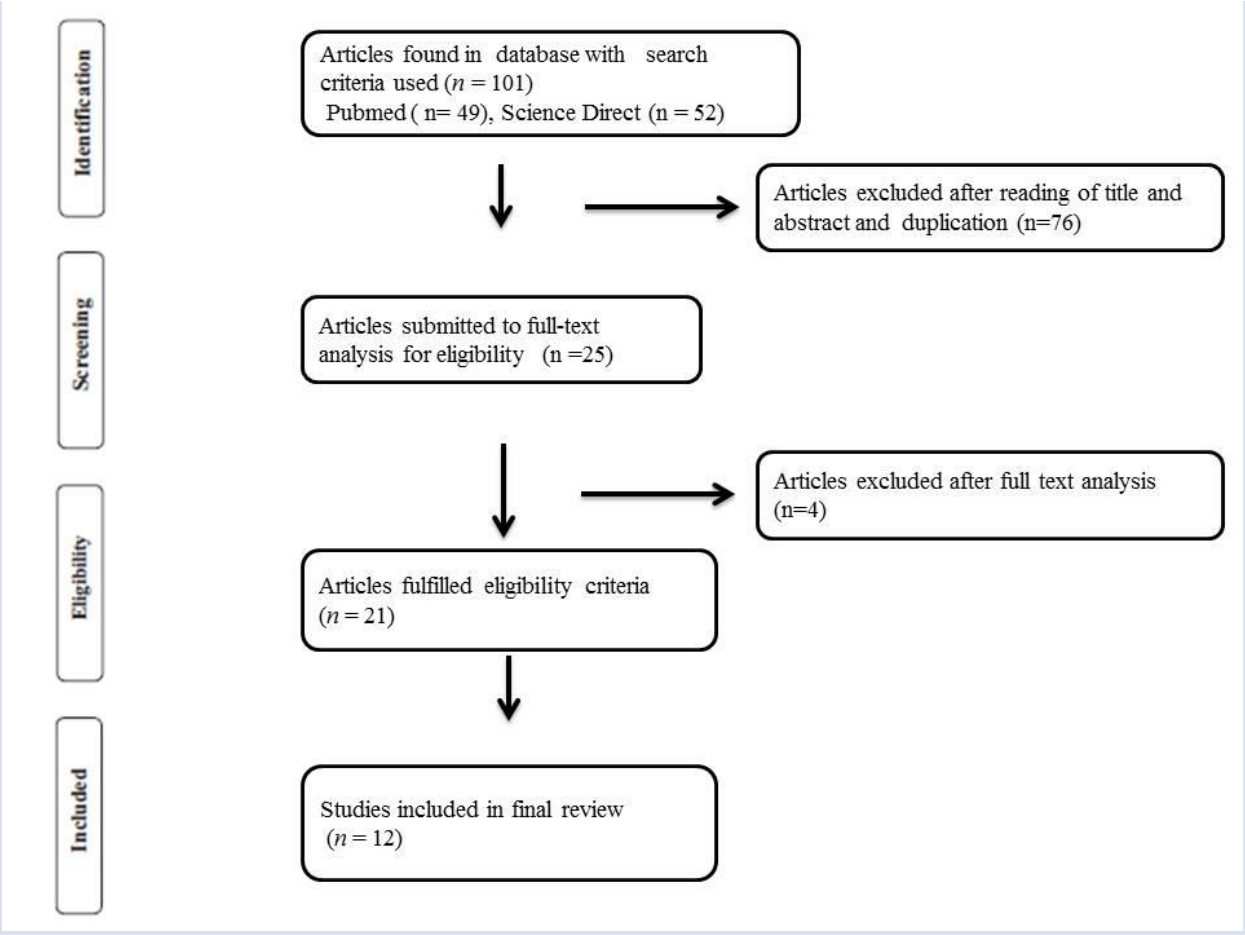

Figure 1. Search Strategy used according to PRISMA statement[8]

Table 1. Summary of Oromandibular dystonia cases reported in the last 10 years with clinical features, treatment protocols used and outcome

SN	Author year	NP	Age and sex	Clinical features	Summary of treatment protocol used	Outcome
1	Schneider R *et al.* (2011)[9]	1	60/Female	Abnormal speech and lisping involuntary movement of mandible, lips, and tongue	Dental prosthesis	Slight improvement of the symptoms but not complete resolution.
2	Jang SM *et al.* (2012)[4]	2	59/ Female 57/ Female	Abnormal jaw protrusive movement after dental extraction, severe dental attrition, unstable occlusion Involuntary jaw tremor, limitation of mouth opening, and protrusive tongue movements following the extraction	Botulinum toxin A injection 30 units per muscle via an extraoral approach Baclofen, analgesics, and occlusal stabilization appliance	Improvement Improvement
3	Bakke M *et al.* (2013)[10]	21	13 Female and 8 Male patients; age range 27-78years with mean age 56.7±12.7 years	Primary and secondary dystonia (13 focal, 7 segmental, 1 multifocal). Problems with mastication and swallowing, hyposalivation, dental attrition, and other dental problems	Intramuscular injection with botulinum toxin Details not reported	Not reported
4	Watt E *et al.* (2013)[3]	1	41/ Male	Involuntary oromandibular contractions	Removable dental appliances - Acrylic block/Splint	Provided beneficial effects
5	Pellecchia MT *et al.* (2014)[11]	1	53/ Female	Feeding difficulties Jaw opening dystonia	Botulinum toxin Injection 80 units of onabotulinum Toxin A in each lateral pterygoid muscle with EMG guidance Trihexyphenidyl (up to12 mg/day).	Effective in relieving jaw opening dystonia Improved tongue protrusion dystonia
6	Khan J *et al.* (2015)[12]	1	77/Female	Involuntary movements of the jaw, Preauricular pain during jaw Function, pain of the mandibular elevator muscles, Persistent twisting of lips, tongue movements	Temporary soft appliance was provided to protect the dentition initially later replaced with hard acrylic appliance 70 units of botulinum neurotoxin lateral pterygoid and orbicularis oris muscles	Reduction of 40% in involuntary Movements Reported complete improvement
7	Van Pelt-Sprangers MJ *et al.* (2015)[13]	1	56/Female	oromandibular dystonia due to capecitabine	Capecitabine discontinued anticholinergic drug	Improvement
8	Pedemonte C *et al.* (2015)[14]	30	Male Patients 18 to 65 years	Post-traumatic oromandibular dystonia bruxism, muscle pain, and involuntary muscle contraction	Use of Onabotulinum toxin A infiltration	Signs and symptoms decreased
9	TA Teemul *et al.* (2016)[15]	6	Female patients 45 to 82 years	Involuntary posterior mandibular movements during mastication, Movements of mandible and tongue, Bruxism, Chronic bilateral dislocation of the TMJ,	Botulinum toxin A 25U to affected masticatory muscles in majority of cases	Improvement
10	Gn S (2017)[7]	1	27/ Female	spontaneous, intermittent, unilateral paroxysmal, severely painful involuntary spasmodic contractions on the right half of face which lasted for 3–5 minutes, repetitive throughout the day	Carbamazepine 200mg BD dose	Complete absence of dystonic movements with improved quality of life
11	Yoshida K (2017)[16]	18	15 Female and 3 Male, mean age: 49.7 ± 16.0 [SD] years, age range: 25to 84 years)	dysarthria, masticatory disturbance, and muscle pain due to prolonged restricted mouth opening	Fifteen patients were treated by injecting botulinum toxin (Botox) into their masseter and temporal muscles. Bilateral coronoidotomy and masseter muscle stripping under general anesthesia combined with muscle relaxation	Overall Improvement in the patients' symptoms of 80.2%
12	Sude A *et al.* (2020)[17]	6	3/Female 3/Male The mean age 62 years (range 52-80 years)	Jaw Pain, teeth attrition, hyposalivation, and masseter hypertrophy	Botox injection affected muscles, Self-care, Physical therapy, Oral appliance, Health psychology	Improvement

SN: Sl Number; NP: Number of patients

Mastication and tongue muscles are most frequently affected in OMD causing involuntary jaw opening/closing movements, jaw deviation, tongue thrusting, lip pursing. Hyperactivity of muscles and masticatory muscle fatigue causes muscle pain. Orofacial region is affected with involuntary movements, soft-tissue trauma, dysphagia and dysphonia. Change in the occlusion and slurring of speech may be observed in some patients. Other symptoms such as excessive drooling, breathing difficulties, and sense of foreign material in the throat were reported. Symptoms are mild initially and become more evident over time.[12] The symptoms may be triggered by certain activities such as talking, mastication and also stress.[1] Due to OMD, damage to dental restorations, dentures, fracture of teeth, excessive dental wear can be seen in patients along with trauma to the lips, gums, and tongue.[1,20] This review revealed involuntary movements, bruxism, jaw pain, masticatory disturbance as common clinical presentation of individuals with OMD.

Bakke *et al.*[10] reported functional and clinical characteristics of 21 OMD cases in 2013. According to them different types of jaw movements were common. They noticed dystonic electromyography activity in anterior digastric muscle (62%) temporal and lateral pterygoid muscles (48%). Issues with mastication and swallowing were commonly found in their study. Other problems associated were hyposalivation, dental attrition, fractured filling, lack of molar support, denture adaptation.[10]

Dystonia can be classified according to etiology, anatomical location and age of onset.[15,20-22]

Classification of dystonia is described in Table 2. Etiology of OMD is not well known.[1] OMD can be classified as primary and secondary in nature. Dystonia is classified as primary if it is inherited or occurs in the absence of other clinical symptoms. Dystonia associated with another known disease is classified as secondary.[19]

The diagnosis of OMD is clinical. Due to its manifestation in various forms and severity, diagnosis of this condition is complicated. Even though this is a rare pathological entity in dental office, the dentist should be familiar with the symptoms to avoid misdiagnosis. A thorough medical history, clinical and neurologic examinations, and investigations such as electromyography is essential for the diagnosis of OMD. Underlying pathology should be ruled out by magnetic resonance imaging of the brain and spinal cord. Preceding the onset of OMD, intermittent, involuntary jaw movements with interference of speech and mastication may be noticed by patients.[18,20]

Clinical examination should be carried out in relaxed position of jaw and during voluntary movement. This is essential as signs of OMD may not be there throughout. Lateral pterygoid, temporalis, masseter and anterior digastric muscles are commonly involved and muscles may be tender on palpation and hypertrophied.[20]

OMD mimics number of dental and medical conditions. Misdiagnosis results in progression of symptoms, incorrect treatment and iatrogenic harm. The differential diagnosis with TMJ disorders (condylar dislocation, bruxism), hemifacial spasm and psychological disorders is necessary. OMD induced bruxism usually stops while sleeping whereas idiopathic bruxism happens during sleep.[20]

OMD can be difficult to discriminate from neurological movement disorders. In Parkinson's disease tremor affects the jaw, mouth or tongue, and can cause clicking of teeth or eating difficulties. Observation of tremor occurrence timing may help to discriminate between the two. Parkinson's exhibits as a "rest" tremor (closed mouth position or open relaxed) and often stops with activities such as mouth opening or talking, whereas voluntary movements usually aggravate OMD.[20] Patients suspected with OMD should be referred to the appropriate health care provider for further evaluation and management.

There is no cure for OMD. Therapeutic management focusses on reducing the dystonic movements, improving patient's aesthetics, masticatory capabilities, swallowing function, and speech. The literature review suggests pharmacotherapy, botulinum toxin type A (BTX) injections, fabrication of occlusal appliances, surgery (peripheral and/or central), chemo denervation, physiotherapy, and occupational therapy as potential treatment options.[1,18,19] Various medications such as anticholinergics, antiparkinson drugs, anticonvulsants, dopamine receptor antagonists, levodopa and lithium were used in the management of OMD.[1]

Table 2. Classification of Dystonia 15,20-22

Classification criteria	Classification subgroup
Anatomical	• Focal: affecting just one part of the body, such as the lower face • Segmental: affecting two contiguous parts such as the lower face and neck • Multifocal: affecting two non-contiguous parts such as the lower face and foot • Generalised: involving the trunk and at least two other sites
Age of onset	• Early onset (infancy, childhood, and adolescence): Mostly secondary dystonia, due to genetic (DYT 1 mutation) or metabolic causes. • Adult onset (21–40 years of age): Idiopathic • Late adult onset (over 40 years of age): mostly idiopathic, few as part of neurodegenerative disorder.
Etiology	• Primary: Idiopathic, Inherited, Familial with genetic predisposition • Secondary: Peripheral trauma, Complication of operation, Diseases of the brain (Neurodegenerative disorders, Cerebral infarction) Drug-induced

Botulinum neurotoxin is the most effective treatment for symptomatic OMDs. Botulinum toxin type A injection is considered as highly effective treatment modality and it is safe. It blocks acetylcholine release at the presynaptic junction which produces transient weakening of the muscle activity. BTX injections have showed 90-95% response rate.[22] Teemul *et al.*[15] reported six patients treated with botulinum A toxin. They used extra oral approach wherein needle was directed through the sigmoid notch. A 40 mm needle of 21-gauge was used and mandibular condyle was identified by palpation on opening the jaw. The needle was inserted to 35-37mm depth from the surface of the skin which is the common position of the lateral pterygoid. They reported that their approach provided benefit for most patients. The injection effect lasted for 7-10 days. Patients were reviewed every three months and injections were repeated every 3-6 months. This systematic review presents the use of botulinum toxin in 8 studies involving 82 patients, which shows that majority of studies used botulinum toxin A for management of OMD. Improvement was noticed in Jang *et al.*[4], Pellecchia *et al.*[11], Pedemonte *et al.*[14], Teemul *et al.*[15], Yoshida[16], Sude *et al.*[17] studies. In Bakke *et al.*[10] study, outcome was not reported. Only Khan *et al.*[12] reported complete improvement. Khan *et al.*[12] first used appliance to protect the dentition which showed 40% reduction in involuntary movement followed by botulinum neurotoxin.

Other treatments include muscle afferent block using intramuscular injection of anaesthetic and alcohol.[22] Pellecchia *et al.*[11] reported a case of drug induced OMD and stated that anticholinergics drugs were effective in relieving tongue dystonia and combination of botulinum toxin injections and anticholinergics were helpful in treating mandibular dystonia.

Dental appliances such as bite block is used for treating OMD. It is custom made for each patient to help stability of jaw and position. Facial appearance, articulatory precision and hyperactive movement's improvement was observed with bite block therapy.[19] Watt *et al.*[3] reported a patient who presented with a 'sensory trick' that is patient symptoms were alleviated by occluding on a pen in between his premolar teeth. Watt *et al.*[3] constructed dental appliances to mimic that effect which was helpful for their patient.

Yoshida[16] suggested coronoidectomy as an effective procedure for patients with severe trismus and ineffective botulinum injection or muscle afferent block therapy. Operative therapies such as peripheral and central nervous system procedures are generally a last resort.[22]

OMD patients may present different clinical conditions to dentists with features of involuntary jaw movements and functional difficulties. Occlusal disturbance and denture retention issues challenge prosthetic treatment. Control of hyposalivation, prevention of dental attrition and fracture of dental restorations due to involuntary jaw movements is essential. Dystonic phenomena complicate dental care, therefore adaption to these special circumstances is required.[12] Jang *et al.*[4] reported two cases of OMD following extraction of lower posterior teeth. It is very important for the dental professional to be familiar with oromandibular dystonia, as it can develop after dental treatment. This condition is often misdiagnosed as a dental problem which may cause significant functional and psychosocial disability.

Conclusions

Oromandibular dystonia is a very rare clinical entity encountered by dental professionals. Diagnosis is very important for the prompt treatment. Since it presents with various forms and different degrees of severity, OMD poses a challenge for dental professionals. Diagnosis requires thorough knowledge and multiple investigations.

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Conflicts of Interest Statement

None.

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