



Prevalence and clinical characteristics of patients with oromandibular dystonia seen in the orofacial pain clinic: a retrospective study

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Objective. The aim of this study was to describe the prevalence and clinical characteristics of patients with oromandibular dystonia (OMD) presenting to an academic orofacial pain clinic. OMD diagnosis was based on clinical signs and symptoms, presenting in varied forms and severities. Knowledge of clinical characteristics would help in early diagnosis and treatment of OMD.

Study Design. In this retrospective study, we identified patients with OMD (International Classification of Diseases [ICD]-9 diagnostic code 333.82 and ICD-10 code G24.4) who presented at the clinic from October 2012 to December 2017. Additional selection criteria were age greater than 18 years and confirmation of OMD diagnosis with at least 1 follow-up visit.

Results. Over a 5-year period, 22 patients with OMD were identified according to the ICD diagnostic codes, and of those, 6 patients met the selection criteria corresponding to a prevalence of 170 per 100,000 (95% confidence interval [CI] 70–390). The most common chief complaint was jaw pain (67%), and all patients had associated diagnosis of myofascial pain of masticatory muscles.

Conclusions. The prevalence of patients with OMD in an academic orofacial pain clinic is higher than previously reported in population-based studies. The presentation of OMD often includes temporomandibular disorders (TMDs), with involvement of various masticatory muscles. (Oral Surg Oral Med Oral Pathol Oral Radiol 2020;130:169–174)

Oromandibular dystonia (OMD) is a form of focal dystonia characterized by involuntary contractions of the muscles of the lower face, jaw, and tongue, and is classified into the following clinical subtypes: jaw opening, jaw closing, jaw deviation, jaw protrusion, lingual dystonia, or combinations of these.^{1–5} OMD can occur alone or can be associated with other forms of focal dystonia, such as blepharospasm, spasmodic dysphonia, or cervical dystonia.^{6,7} It can be primary (idiopathic) or secondary to encephalitis; ischemia; metabolic disorders, such as Wilson disease; or neuroleptic medications (chlorpromazine, haloperidol, etc.). Secondary causes are often ruled out on the basis of blood workup, neuroimaging findings, and patient history of neuroleptic medication use.^{8,9} Peripheral injury to tissues, such as orofacial trauma from ill-fitting dentures or dental extractions, have also been reported to trigger the onset of OMD.⁷ It interferes with jaw functions, including chewing, swallowing, and talking. Furthermore, it could result in social embarrassment and impaired quality of life.^{3,7} Patients often develop unique tactile stimulation techniques well known as “sensory tricks,” such as

touching the face, bending the neck forward, singing, humming, yawning, or holding an object between the teeth for temporary alleviation of dystonic movements.^{10,11} Management of OMD primarily includes botulinum toxin injections to the affected masticatory muscles, oral medications (tetraabenazine, baclofen, clonazepam), and oral appliances.¹²

OMD is a clinical diagnosis; therefore, heterogeneity in clinical presentation and lack of clinical guidelines pose a significant challenge to the clinicians in early diagnosis. The average delay from symptom onset to diagnosis of focal dystonia ranges from 4.8 years to 10.1 years.¹¹ Familiarity with the clinical characteristics of OMD would help in its early diagnosis, and in this regard, several case reports have described the dentist’s point of view in diagnosing this rare movement disorder.^{13–17} Given the orofacial presentation of OMD and the involvement of the masticatory muscles, exploration of the prevalence and clinical characteristics in patients presenting to the orofacial pain clinic seems warranted. Hence, the aim of this study was to assess the prevalence and clinical characteristics of patients with OMD presenting to an orofacial pain clinic at the University of Minnesota (Minneapolis, MN).

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Received for publication Jan 15, 2020; returned for revision Apr 6, 2020; accepted for publication May 4, 2020.

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2212-4403/\$-see front matter

<https://doi.org/10.1016/j.oooo.2020.05.002>

Statement of Clinical Relevance

The clinical features of oromandibular dystonia can include involuntary movements, along with comorbid temporomandibular disorders or jaw pain. Dentists should be familiar with the clinical signs and symptoms of oromandibular dystonia for timely diagnosis.

MATERIALS AND METHODS

We conducted a retrospective study at the TMD, Orofacial Pain, and Dental Sleep Medicine Clinic at the University of Minnesota (Minneapolis, MN). In this academic orofacial pain clinic, patients are seen by residents under the supervision of board-certified orofacial pain specialists. The study protocol was approved by the local institutional review board, and the requirement for informed consent was waived. Patients diagnosed with OMD were identified from the electronic dental record system “Axium.” Inclusion criteria were age greater than 18 years, a diagnosis of OMD, during the period October 2012 to December 2017, with either International Classification of Diseases (ICD)-9 diagnostic code 3382 or ICD-10 diagnostic code G24.4. The selected timeframe was used on the basis of the implementation of the electronic dental record system at our institution and the institutional review board protocol. Another key inclusion criterion was confirmation of OMD diagnosis on at least 1 follow-up visit to ensure that the OMD diagnosis was an established one, rather than a provisional one. Furthermore, in addition to the ICD diagnostic codes, the individual charts of eligible patients were reviewed to confirm the accuracy of the documented diagnosis. All of the patients in our clinic were ultimately evaluated and diagnosed by board-certified orofacial pain specialists. A detailed review of the patients’ charts was performed to access all necessary information, including demographic data (age, gender, ethnicity); clinical features (chief complaint, onset, duration, frequency, aggravating and alleviating factors, associated symptoms including temporomandibular disorder [TMD]–related symptoms); presumed etiology; medical, social, and psychosocial histories; diagnoses based on clinical examination (extraoral examination, including type of movement observed, facial features, temporomandibular joint [TMJ] noises, pain to palpation and intraoral examination, including range of motion, occlusal wear, hyposalivation); number of providers seen; and recommended treatments. Descriptive statistics were used to summarize the data. The continuous variables were presented with mean and range (minimum–maximum), and categorical variables were presented with counts and percentages.

RESULTS

Over a 5-year period, 3572 new patients were evaluated in the TMD, Orofacial Pain, and Dental Sleep Medicine Clinic at the University of Minnesota (Minneapolis, MN). Twenty-two patients were identified by using the ICD diagnostic codes for OMD. Of these patients, only 6 patients met the inclusion criteria with a confirmed diagnosis of OMD on at least 1 follow-up visit (Figure 1). This translates to a clinical prevalence of 170 per 100,000 persons (95% confidence interval [CI] 70–390) for OMD in our clinic population.

Of the 16 patients who were excluded, 5 patients were diagnosed with OMD at the initial visit but were referred to a movement disorder clinic; no follow-up visits at our clinic were required. These 5 patients were referred to the OMD clinic because of the complexity of their management, as determined by the respective provider. Two patients came to the clinic with a diagnosis of OMD but did not have follow-up visits to confirm the diagnosis; therefore, they did not meet the selection criteria. One patient was younger than 18 years of age, and 1 case was outside the prespecified observation window for the review of records. Of the remaining 7 patients who were excluded, the OMD diagnosis could not be confirmed during the follow-up visits. They were diagnosed with atypical facial pain, trigeminal neuralgia, tardive dyskinesia, or hemifacial spasm. Given the clinical diagnosis of OMD, radiologic examination was performed, when deemed necessary by the provider, to rule out any secondary diagnoses.

The mean age of the study patients was 62 years (range 52–80 years), and 3 patients (50%) were females. Patients’ clinical characteristics are presented in Table I. The most common chief complaint was jaw pain (4 of 6 [67%]). All 6 patients with OMD received a diagnosis of myofascial pain of masticatory muscles, and 67% patients received a diagnosis of TMJ arthralgia. Other commonly noted clinical features were teeth attrition (4 of 6 [67%]), hyposalivation (2 of 6 [33%]), and masseter hypertrophy (2 of 6 [33%]) (Table II) and were assessed objectively by their clinicians. Psychosocial comorbidities were present in 5 of 6 patients (83%); their conditions included anxiety, depression, or both. The sensory trick was evident only in 1 patient (17%); it involved putting a finger between his front teeth. In addition to OMD, these patients were also diagnosed with TMJ degenerative joint disease ($n = 2$; 33%) based on radiographic findings of cone beam computed tomography (CBCT) and internal disk displacement ($n = 1$; 17%). The average number of providers seen by an individual OMD patient was 5 (range 2–12). The commonly seen providers included orofacial pain specialists, movement disorder specialists, dentists, health psychologists, and physiotherapists.

DISCUSSION

In our study, we found the prevalence of OMD to be 170 per 100,000 persons (95% CI 70–390), which is much higher compared with the previously published community-derived population reports. Prevalence of OMD in a Minnesota community population was estimated to be 6.9 per 100,000.¹⁸ Similarly, other studies reported OMD prevalence to be 2.8 per 100,000 in Iceland and 2.1 per 100,000 in the Faroese population.^{19,20} The high prevalence noted in our study likely resulted from the high level of suspicion of OMD in the clinicians

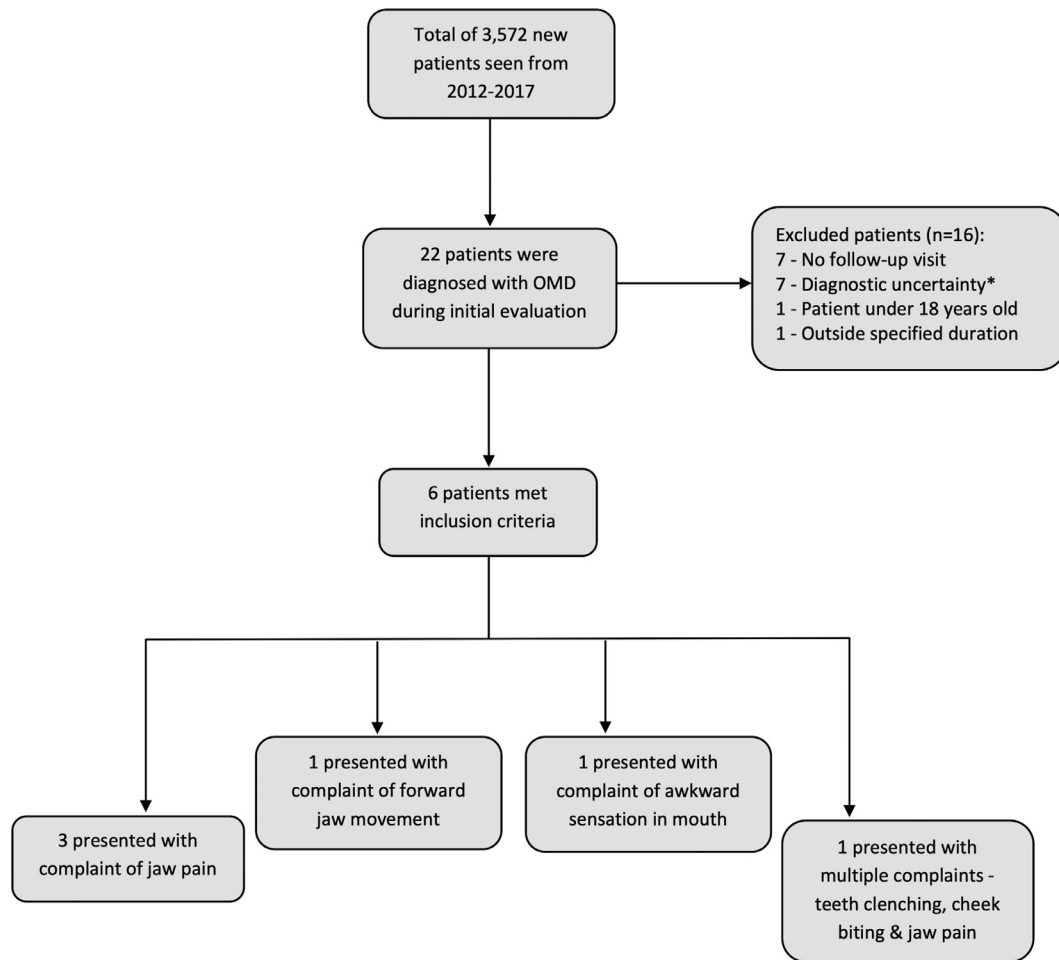


Fig. 1. Flow chart representing the patient selection. *Patients had diagnosis other than oromandibular dystonia (OMD) that included atypical facial pain, trigeminal neuralgia, tardive dyskinesia, and hemifacial spasm.

in our orofacial pain clinic or a possible association of OMD to TMD leading to a high proportion of patients with OMD visiting the orofacial pain clinic. The mean age at onset of OMD in our study was consistent with that reported by other population studies. OMD is reported to be 4 times more prevalent in females, but we observed an equal distribution of OMD in both genders.¹⁸ This most likely resulted from the small sample size in our study. We did not find specific clinical features, such as facial grimacing, lip pursing, lip smacking, and tongue protrusion, in our study, which are more commonly seen in patients with tardive dyskinesia.³

Use of a sensory trick was noted in only 1 patient. Studies have shown that the use of an oral appliance to function as a sensory trick could lead to minimal to partial resolution of symptoms.^{10,13,15,21} According to our patient's report, the stabilization splint helped, for a short duration, reduce dystonic activity, and the patient continues to place his finger between his teeth to obtain temporary relief. This shows that sensory tricks decrease the abnormal facilitation of dystonic

activity in the brain but that this effect will diminish as the nervous system becomes accustomed to it over a period.²² The sensory trick can add specificity to the diagnosis of OMD, as suggested by Defazio et al.²³

In our study, 5 of 6 patients had comorbid psychosocial conditions. Previous studies have reported that patients with craniocervical dystonia, including OMD, are susceptible to behavioral disorders. However, whether psychosocial disorders have a causal relationship to OMD or are a secondary effect of social and physical disfigurement that occurs in OMD remains unknown.^{2,24}

Jaw pain in OMD can be thought of as a result of long-lasting dystonic muscle contraction. In fact, it was the most common presenting complaint that caused 4 of 6 OMD patients to seek care in our clinic. It was interesting to note that all 6 patients in our study had an associated diagnosis of myofascial pain of masticatory muscles, and 4 had TMJ arthralgia. None of the study patients had a diagnosis of other subtypes of myalgia, that is, local myalgia (localization of pain only at the site of palpation) or myofascial pain with

Table 1. Baseline demographics, clinical characteristics and treatment of patients with OMD

Patient	Age/Gender	Type of OMD	Chief complaint	Other clinical features	Functions affected	Sensory trick	Etiology	Secondary diagnoses	Treatment	Number of providers seen at the time of diagnosis
1	62/M	Jaw protrusion	Forward jaw movement and jaw tightness	Jaw pain, stuttering	Speech, smiling	Yes; keeping finger between teeth	Idiopathic	Myofascial pain, masseter hypertrophy	Botox injection in bilateral lateral pterygoid muscles	6
2	52/M	Unknown	Awkward sensation in mouth and affected function of the tongue	Difficulty swallowing	Speech	None	Idiopathic	Myofascial pain, TMJ arthralgia, DJD	Self-care, physical therapy, oral appliance, health psychology	3
3	72/M	Jaw deviation	Jaw pain	Jaw deviation to the right side	None	None	Stroke	Myofascial pain, TMJ arthralgia, Internal derangement	Botox injection in lateral pterygoid muscle	2
4	53/F	Jaw closing	Clenching, prominent jaw muscles, cheek biting, jaw pain, headaches	None	None	None	Idiopathic	Myofascial pain, TMJ arthralgia, tension-type headaches	Botox injection in bilateral masseter muscles	2
5	80/F	Jaw closing	Jaw pain	Uncontrollable clenching	None	None	Idiopathic	Myofascial pain, TMJ arthralgia	Referred to movement disorder specialist	4
6	53/F	Mixed - Lingual dystonia and jaw deviation	Jaw pain with chewing and grinding	None	Eating	None	Idiopathic	Myofascial pain, DJD	Botox in lateral pterygoid muscle, self-care, physical therapy, oral appliance, health psychology	12

DJD, degenerative joint disease; *OMD*, oromandibular dystonia; *TMD*, temporomandibular disorder; *TMJ*, temporomandibular joint.

Table II. Frequency of clinical characteristics of patients with oromandibular dystonia presenting to our clinic

Clinical features	Frequency
Jaw pain	67%
Dental attrition	67%
Hyposalivation	33%
Masseter hypertrophy	33%

referral (referral of pain beyond the boundary of the muscle being palpated). The findings in our study are similar to those of the study by Costa et al., who attempted to assess the relationship between TMD and craniocervical dystonia and found that all 20 patients with OMD had some form of TMD.²⁵ Yoshida et al. reported that 7 of 385 patients in the OMD group had secondary TMJ symptoms.²⁶ Slaim et al. reported pain in 32% of patients with OMD, but their report lacked further specifications of pain and Sinclair et al. reported that 34% patients experienced pain.^{5,27} All these studies indicate that pain and coexisting TMD are often associated with OMD and that clinicians should screen patients with OMD for TMD because this may influence patient outcome.^{5,26,27}

The findings of our study need to be interpreted in light of a few limitations. First, this is a retrospective study that relied on accurate and complete documentation. To overcome this limitation, clinicians should focus on detailed documentation of OMD cases. Second, the study sample is small, given the lower overall prevalence of OMD and the limited number of patient seen in a single clinic setting. Also, it is not uncommon to miss eligible patients during the transition phase between stages in the ICD coding system. We attempted to minimize this limitation by using both ICD 9 and ICD 10 diagnostic codes that covered our entire study period. Finally, there could be a referral bias because patients with OMD and TMD are more likely to be referred to an orofacial pain clinic. This referral bias could also have resulted in the higher prevalence of OMD noted in our study because a specialty clinic in an academic setting is more likely get referrals compared with community practices. Future studies are needed to compare the clinical characteristics of patients presenting to an orofacial pain clinic with those of patients presenting to a neurology or movement disorder clinic.

CONCLUSIONS

Our study found an OMD prevalence of 170 per 100,000 persons in our academic orofacial pain clinic setting, which is higher than that reported in previous studies with community populations. All of the patients with OMD in our study had an associated diagnosis of

some form of TMD, which raises the question “Do patients with OMD, in general, have TMD?” Dentists need to be familiar with the clinical characteristics of OMD because they will, at some time or another, be encountering patients with this disorder.

PRESENTATION

Preliminary results of this study were presented at the 42nd scientific meeting of the American Academy of Orofacial Pain, April 26–29, 2018, held in Chicago, IL, as a poster presentation.

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