

Clinical Features of Oromandibular Dystonia

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Oromandibular dystonia (OMD) is a form of focal dystonia that affects the masticatory, facial and lingual muscles in any variety of combinations, which results in repetitive involuntary and possibly painful jaw opening, closing, deviation or a combination of these movements. This study aimed to investigate clinical features and treatment type of OMD patients.

By retrospective chart review, the study was conducted to consecutive OMD patients who visited a department of Oral Medicine and Orofacial Pain Clinic in a university dental hospital during Aug 2007 to Apr 2010.

78 OMD patients were identified with female preponderance (M:F=1:3.6) and a mean age of 72 years. Their mean duration of OMD was about 10 months. The most common chief complaints at the first visit was jaw ache, followed by uncontrolled, repetitive movement of the jaw and/or oral tissues, pain in the oral region ($p=0.000$). The most common subtype of OMD was lateral jaw-deviation dystonia, followed by combination and jaw-closing dystonia ($p=0.001$).

While no apparent cause was recognized in over 60% of the OMD patients, peripheral trauma including dental treatment such as prosthetic treatment and extraction was the most frequently reported as precipitating factor ($p=0.000$). Medication was the 1st line therapy for our patients and anxiolytics such as clonazepam was given to most of them.

Based on the results of this study, OMD is the disease of the elderly, particularly of women and causes orofacial pain and compromises function of orofacial region. Some patients considered dental treatment a precipitating factor. Dentists, therefore, should have knowledge of symptoms and treatment of OMD.

Key words : dystonia, oromandibular dystonia, dental treatment

I. INTRODUCTION

Oromandibular dystonia, OMD is a form of focal

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dystonia that affects the masticatory, facial and lingual muscles in any variety of combinations, resulting in repetitive and sometimes sustained jaw opening, closure, deviation or any combination of these.^{1,2)} Compared to dyskinesia, OMD is intermittent and present as short, sustained muscle contractions resulting in abnormal muscle movements and posturing.³⁾

The prevalence of OMD was estimated to be approximately 3 to 30 per 100,000 persons in the United States^{4,5)} and OMD affects more women than men with a mean age of symptoms onset between 31 and 58 years.^{2,6)}

OMD can be idiopathic, tardive (drug-induced) or secondary to other movement or neurological disorders.⁷⁾ Genetic predisposition may be considered in some cases.⁸⁾ Tan and Jankovic²⁾ indicated that most OMDs (63%) are idiopathic in etiology. Tardive dyskinesia, a variant of tardive dyskinesia, should be considered when there is a history of neuroleptic exposure during at least 6 months before the onset of movement disorder.⁹⁾ The prevalence rate of tardive dyskinesia is found to be about 10% in a psychiatric population.¹⁰⁾ Some researchers mentioned dental factor including edentulousness and placement of ill-fitting dentures as precipitating factor for symptom onset although the mechanisms remain ill-defined.^{11,12)}

Pathophysiology for OMD is unknown, however, it probably has multifactorial mechanisms involving basal ganglia dysfunction, hyperexcitability of motor neurons involved in signaling, reduced inhibition of spinal cord and brain stem signals coming from supraspinal input, and dysfunction of neurochemical systems involving dopamine, serotonin and noradrenaline.^{5,13)}

OMD results in a variety of complications such as tooth wear, tooth and denture damage, accelerated bone loss in edentulous patients, oral pain, temporomandibular joint (TMJ) degeneration, mandibular luxation, friction/biting injuries (tongue, cheek bites), which subsequently lead to speech impairment, dysphagia, chewing difficulties, inadequate food intake and weight loss, and displacement/impaired retention of removable dental prostheses. In addition to these physical and functional disability, OMD may cause social embarrassment including unemployment, isolation, depression and etc.¹⁴⁾

OMD is often misdiagnosed, primarily because of the numerous types and severities in the presentation of OMD. There is no available gold standard for diagnosis of different types of OMD as yet. Misdiagnosis commonly includes TMD or spontaneous condylar dislocation, hemimasticatory or hemifacial spasms and psychologic manifestation. Inappropriate diagnosis subsequently leads to

improper management.³⁾

For improvement of diagnosis and management, clinical features and treatment for OMD patients still need to be well-defined. Besides, it is difficult to find the researches about Korean OMD patients. Therefore, this study aimed to investigate clinical features such as age, gender, subtype of OMD, duration of symptom, precipitating factor of symptom onset, comorbidity for the OMD patients attended to Orofacial Pain Clinic in a university dental hospital in Korea. Type of their treatment were also evaluated.

II. SUBJECTS AND METHODS

Retrospective chart review was conducted to consecutive OMD patients who visited a department of Oral Medicine and Orofacial Pain Clinic in a university dental hospital (Dankook University Dental Hospital, Cheonan, Korea) during Aug 2007 to Apr 2010. Exclusion criteria was those with incomplete clinical records. The study plan was approved by the ethic committee of the same hospital (IRB No. H-1004/002/005).

Data collected was as follows; gender, age, chief complaint, duration of symptom, cause or precipitating factor, comorbidity and medication, subtype of OMD, oral condition (status of dentition and oral mucosa, presence or absence of pain), and type and outcome of treatment applied. Status of dentition was divided into 4 categories; complete denture (those with upper and lower CDs), removable partial dentures, extensive prosthesis (those with fixed prosthesis > 3-unit bridges) and full dentition. Subtype of OMD was divided into jaw-closing, jaw-opening, jaw-deviation, lingual and combination dystonia.

Type of OMD treatment was divided into 3 categories; medication, botulinum toxin type A injection, oral appliance therapy. Medication therapy included anxiolytics (clonazepam, diazepam), muscle relaxants (baclofen, eperisone), analgesics, tricyclic antidepressants(TCA), anticonvulsants (gabapentin, pregabalin).

Statistical analysis was done using SPSS ver 14.0. To clarify demographics and clinical features for OMD patients, chi-square tests were performed. Level of significance was defined as $p < 0.05$.

III. RESULTS

78 OMD patients were identified with preponderance of female gender (M:F=1:3.6) and a mean age of 72.3 ± 9.7 years (range: 34 years to 88 year) in this study. Their mean duration of symptom was about 10.7 ± 14.4 months (max: 6.5 years).

The most common chief complaint of them at the first visit was jaw ache (56%), followed by uncontrolled, repetitive movement of the jaw and/or oral tissues (25.6%), pain in the oral region such as teeth, gingivae and other soft tissues, difficulty in chewing or swallowing and occlusal instability

factors related to onset of OMD, no apparent cause ($p=0.000$, Table 1). In regards with precipitating was recognized in the most of cases (66.7%). On the while, dental treatment including placement of prosthesis and extraction was the most commonly considered as precipitating factor. Trauma, medication, and emotional event were also reported ($p=0.000$, Table 1).

Table 2 represents comorbidity and medication of the OMD patients. 87.2% of them (N=68) were suffering from systemic diseases; cardiovascular diseases including hypertension (51%), psychiatric disorders such as depression (24%), neurological diseases (20%) and diabetes (15%). 60% patients had TMD ($p=0.000$). The OMD patients were taking cardiovascular drugs (59%), neurological drugs (21%), antidepressants (19%), anxiolytics (10%) ($p=0.000$, Table 2)

Table 1. Chief complaints and precipitating factors for the OMD patients.

<i>Chief Complaints</i>		
Jaw ache	N=44 (56.4%)	
Uncontrolled movement of the jaw and/or the oral tissues	N=20 (25.6%)	
Pain in the oral tissues (oral soft tissue, gingivae, tooth)	N=11 (14.1%)	$\chi^2=59.480,$ $p=0.000$
Difficulty in chewing or swallowing	N=10 (12.8%)	
Occlusal instability, discomfort for prosthesis	N=4 (5.1%)	
Others (oral soft tissue trauma, burning mouth, dry mouth, etc)	N=11 (14.1%)	
<i>Precipitating factors</i>		
No apparent cause	N=52 (66.7%)	
Dental treatment	N=15 (19.2%)	
Trauma	N=5 (6.4%)	$\chi^2=59.480,$ $p=0.000$
Medication	N=4 (5.1%)	
Emotional event	N=2 (2.6%)	
<i>Chi-square tests</i>		(N=78)

Table 2. Comorbidity and medication of the OMD patients

<i>Comorbidity</i>		
TMD	N=47 (60.4%)	$\chi^2=25.301$ $p=0.000$
Cardiovascular disorders	N=40 (51.3%)	
Psychological disorders	N=19 (24.4%)	
Neurologic disorders	N=16 (20.5%)	
Diabetes Mellitus	N=12 (15.4%)	
Others	N=12 (15.4%)	
<i>Types of medication</i>		
Cardiovascular drugs	41 (58.6%)	$\chi^2=45.229$ $p=0.000$
Neurologic drugs	15 (21.4%)	
Antidepressants	13 (18.6%)	
Anxiolytics	7 (10.0%)	
Others	21 (30.0%)	
<i>Chi-square tests</i>		

Table 3. Subtype of OMD.

Lateral jaw deviation dystonia	N=26 (33.3%)
Jaw closing dystonia	N=25 (32.1%)
Combination dystonia	N=23 (29.5%)
Lingual dystonia	N= 4 (5.1%)
Jaw opening dystonia	N= 0 (0%)
<i>Chi-square test, $\chi^2=16.667, p=0.001$</i>	
(N=78)	

As seen in Table 3, lateral jaw-deviation dystonia was the most common subtype of OMD and combination and jaw-closing dystonia were also frequent ($p=0.001$, Table 4). While patients presenting with lingual dystonia only was uncommon (5%), lingual dystonia was frequently observed in combination with any of others (65.2%). A single case revealed jaw opening dystonia in combination of jaw opening type.

Oral conditions for the OMD patients were presented in Table 4. Those with extensive prosthesis, i.e., fixed prosthesis of > 3-unit bridges, were the most common in our OMD patients (46.8%,

$p=0.000$). Traumatic oral ulcer was found in 18% of the OMD patients and 85% reported pain or discomfort ($p=0.000$, Table 4).

Medication was given as the 1st-line therapy to all our patients and 97% of them were prescribed with anxiolytics such as clonazepam. Analgesics, muscle relaxants, TCAs, anticonvulsants were also frequently given ($p=0.000$, Table 5). Botulinum toxin injection and oral appliance were performed in 4 and 3 cases, respectively.

IV. DISCUSSION

This study investigated patients attending to orofacial pain clinic in dentistry and the demographic results are consistent with results of previous studies.^{2,4,5} OMD is a disease of elderly people with female preponderance (mean age of 72.3±9.7 years, M:F=1:3.6) The most common chief complaint for our OMD patients was jaw ache rather than uncontrolled jaw movement (Table 1), which may in part be explained by that TMD was frequently comorbid (60.4%, Table 2) and that OMD was often noticed by practitioners.

Table 4. Oral condition of the OMD patients.

<i>Status of dentition</i>		
Extensive prosthesis (> fixed prosthesis of 3 unit bridge)	N=37 (46.8%)	
Full dentition (≤ fixed prosthesis of 3 unit bridge)	N=21 (26.6%)	$\chi^2=31.641,$ $P=0.000$
Removable partial dentures	N=2 (2.6%)	
Complete dentures	N=18 (23.1%)	
<i>Presence of pain/discomfort</i>		
Yes	66 (84.6%)	$\chi^2=37.385$ $P=0.000$
No	12 (15.4%)	
<i>Presence of traumatic ulcer</i>		
Yes	14 (17.9%)	$\chi^2=32.051$ $P=0.000$
No	64 (82.1%)	
<i>Chi-square tests</i>		(N=78)

Table 5. Type of OMD treatment applied.

<i>Medications</i>	78 (100%)
Anxiolytics (clonazepam, diazepam)	76 (97.4%)
Muscle relaxants (baclofen, eperisone)	33 (42.3%)
Analgesics	39 (50.0%)
TCA	21 (26.9%)
Anticonvulsants (gabapentin, pregabalin)	17 (21.8%)
<i>Botulinum toxin A injection</i>	4 (5.1%)
<i>Oral appliance therapy</i>	3 (3.8%)
<i>Chi-square test, $\chi^2=138.539, P=0.000$</i>	
<i>TCA: tricyclic antidepressants.</i>	
(N=78)	

Etiology and pathophysiology of OMD has not been fully understood. In a study by Tan and Jankovic,²⁾ most OMDs are idiopathic in etiology (primary dystonia), accounting for 63% of cases reported. Similar finding was also observed in this study; 67% of the OMD patients were not aware of any apparent cause (Table 1). Dystonia (secondary dystonia) may develop due to environmental factors such as peripheral trauma and after medication. Tan

and Jankovic²⁾ also reported; tardive OMD (22.8%) and peripheral-induced OMD (9.3%).(G) The common form of all secondary dystonia is tardive dyskinesia, which develops as a side effect of long-term treatment with antipsychotic drugs (haloperidol, clozapine, risperidone).¹⁵⁾ Other offending drugs include antiemetics (metoclopramide, promethazine), antiparkinsonian agents (levodopa, benztropine, trihexyphenidyl), anticonvulsants (phenytoin,

carbamazepine), antihistamines (diphenhydramine, ranitidine), tricyclic antidepressants (amitriptyline, doxepin), selective serotonin reuptake inhibitors (SSRI; fluoxetine, paroxetine, sertraline).^{3,14,16} Patients with long-term use of any of drugs was 89.7% in this study (Table 2).

Some cases of OMD after dental treatment have been reported, although the causal relationship between these procedures and dystonia is still unclear¹⁷. Sankhla et al¹⁸ reported 27 peripherally induced OMD, four of which were wearing new sets of dentures, including one patient with an ill fitting dental bridge. The patient with ill fitting dentures had a history of using their jaw muscles to manipulate their jaw position to adapt to the new dentures. These malaligned dentures may have caused a proprioception impairment of the oral cavity leading to subsequent development of dystonia or so-called 'edentulous dyskinesia'.

20% of the OMD patients in this study considered any relation of dental treatment with OMD onset, most of which included prosthetic treatment such as placement of bridge or denture (Table 1). Invasive dental treatment such as implant surgery and extraction was also mentioned in some cases (2 and 4 patients of them, respectively) Although the causal relationship between dental treatment and OMD has not been conclusive, dental treatment may enhance patients' awareness to their facial and/or oral structures at any rate. 6.4% of the patients recognized peripheral trauma including surgery as a precipitating factor of OMD, which means that total 25.6% of 78 OMD patients were thought to be peripherally-induced in etiology.

There have been a few studies concerning about frequency of OMD subtype. Lateral jaw-deviation (33%) and jaw closing (32%) dystonia was the most common subtype of our 78 patients with OMD (Table 3). Combination dystonia was found in 29.5% and lingual dystonia in 22.4% (N=19). It is of interest that lingual dystonia coexisted frequently with any of other movement in combination dystonia (65.2% of 23 combination cases) while a single case exhibited lingual dystonia only. Tan et al¹⁹

investigated 116 OMD patients and reported that jaw closing dystonia was the most frequent (49.1%). Lingual dystonia in their study was found in 19.8% of the their cases. They also indicated that oro-facial-lingual stereotypes were more frequently in the tardive than the idiopathic dystonia.

Although there has been no standard guideline for treatment of OMD, a three-step process is suggested for the treatment.²⁰ The first step is to collect a full clinical history and examination for ruling out CNS disease, adverse medication reactions and local pathology. The second step is to try motor-suppressive medication including anticholinergics, benzodiazepines, antiparkinsonism drugs, anticonvulsants, baclofen and etc. The third step is to consider botulinum toxin (BTX) injections if the disorder is focal and medication are proven to be ineffective.

The first line therapy for all of our patients was medication and clonazepam was prescribed in 97% cases. Baclofen was also frequently tried. Frequent usage of muscle relaxants and analgesics was due to TMD pain which coexisted in some OMD patients and in part because OMD was misdiagnosed as TMD. There was no evidence-based information about the efficacy of the different pharmacologic therapeutic options being applied in dystonia.²¹ Systemic pharmacological therapy benefits about one third of patients²² and anticholinergics have been found to be the most effective oral medication for the treatment of dystonia.²³

When medication was not effective enough or ineffective at all, BTX injection is recommended. Numerous studies have confirmed a 90-95% response rate to BTX injections.²⁴ In a comparative study between jaw-closing and jaw-opening dystonia by Singer et al¹¹ reported that better response to BTX injections were more frequent in jaw-closing dystonia while patients with jaw-opening dystonia were more likely to experience benefit from oral antidystonic medications.

Oral appliance tried in a few cases was not to relieve OMD symptoms but to protect oral tissues from frequent trauma due to aberrant repetitive

dystonic movement of the jaw and/or tongue.

This study has some limitation. It was not easy to get information about drug history, mainly because a number of patients were not aware of what specific kind of drug they were taking for their systemic disease. It is thought that doctors should give more written-information about treatment including medication to their patients.

In addition, use of valid rating scale with long-term follow-ups and evaluation about patients' satisfaction should be required for better assessment of treatment outcome.

The diagnosis of OMD is clinical and complicated, since it can present in various forms and with different severity.¹⁷⁾ It's not easy to clear cause or precipitating factors for development of OMD. Comprehensive and standardized clinical evaluation using a OMD-specific structured chart form would be helpful for diagnosis and management of OMD.

OMD is a disease difficult to manage and may cause considerable functional and psychosocial disability. It is often misdiagnosed as a 'dental problem' or may be developed or recognized after dental treatment. Dentists, therefore, should have knowledge of symptoms and management of OMD. A comprehensive study with larger sample needs to be performed to compare clinical features and treatment outcomes related to OMD subtype.

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국문초록

하악운동이상증의 임상양태

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하악운동이상증(oramandibular dystonia, OMD)은 턱이나 구강조직의 반복적, 불수의적, 비정상적인 움직임으로 정의되며 저작기능과 교합에 문제를 초래하여 치과치료를 어렵게 하고 보철물의 유지근란을 야기할 뿐 아니라 잦은 구강 연조직 손상이나 만성 안면통증의 원인이 되기도 하므로 치과의사의 관심이 필요한 질환이다. 본 연구는 단국대학교 치과대학 부속치과병원 구강내과에 내원하여 OMD로 진단되어 치료를 받은 환자들의 병력기록부상의 관련 항목을 조사하여 OMD 환자들의 임상적 특징과 치료 유형을 분석하였다.

2007년 8월부터 2010년 4월까지 단국치대 부속병원 구강내과에 내원한 78명의 OMD 환자는 평균 72세의 고령이었으며 여자가 남자에 비해 3.6배 많았고, 평균 10개월 이상의 비교적 긴 병력기간을 보였다. 내원한 OMD 환자의 가장 흔한 주소는 턱의 통증이었으며 그 다음으로 빈도가 높은 주소는 턱과 구강조직의 조절 안 되는 반복적인 움직임과 구강조직의 통증이었다($p=0.000$). OMD 유형은 폐구형(jaw-closing dystonia)이 가장 많았고, 측방운동형(lateral jaw-deviation dystonia)과 복합형(combination dystonia)도 많이 발생하였다($p=0.001$). 환자의 60% 이상은 OMD 발생과 관련하여 뚜렷한 원인을 인지하지 못했으나 약 19%의 환자들은 보철치료, 발치 등의 치과치료와 관련 있다고 보고하였다($p=0.000$). 본원에 내원한 환자는 약물치료가 가장 먼저 시도되었고 진정제(clonazepam)가 거의 모든 환자(97%)에서 처방되었다.

이상의 결과로 볼 때, OMD는 뚜렷하게 노인, 특히 여성에게 많이 발생하는 질환으로서 턱과 구강조직의 통증을 유발하며 과반수 이상은 뚜렷한 원인을 모르지만 일부 환자들은 치과치료 후에 자신의 문제를 처음 인지하는 경향이 있다. 그러므로 치과의사는 환자를 치료할 때 OMD의 존재를 간과하지 않도록 관심과 주의를 기울일 필요가 있다.

주제어 : 운동이상증, 하악운동이상증, 치과치료