









RESEARCH ARTICLE

OROMANDIBULAR DYSTONIA: PREVALENCE, CLINICAL AND DEMOGRAPHIC DATA, THERAPEUTIC STRATEGIES OUT-COME FOR HUNDRED PATIENTS IN SANA'A CITY, YEMEN

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Abstract

Background and aims: Dystonia is a movement disease characterized by persistent, uncontrollable muscle contractions that often lead to abnormal postures. The tongue, jaw, and mouth are all affected by OMD, which is an uncommon focal dystonia. OMD is a rare public health problem that takes time to diagnose and treat because it is frequently identified as psychogenic. This study sought to ascertain the frequency, demographics, clinical features, and treatment outcomes of patients with mandibular dystonia (OMD) in Sana'a, Yemen.

Methods: A prospective cross-sectional study was conducted; the study included 100 Yemeni adults attending the Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Sana'a University Clinics and Departments of Oral, Maxillofacial and Neurosurgery at Kuwait University Hospital and some private centers for maxillofacial medicine and neurosurgery in the city of Sana'a during the year 2022. The selected patients were complaining of contraction or involuntary movement of the masticatory, lingual and/or lower facial muscles. Age, gender, age at onset of illness, duration of illness, personal habits, type of OMD, previous and current treatments, and their outcomes were recorded on a standard, pre-designed questionnaire.

Results: The incidence rate of OMD is 19/100,000/year. Most of the patients (92%) are male while only 8% are female. The first episode onset is after stress (29%), 11% after illness, 6% after dental treatment, and 48% without causes. Jaw deviation dystonia was dominant at 65%, followed by jaw closing dystonia (42%), jaw protrusion dystonia (29%), and jaw opening dystonia (27%). After current treatment out-come, there was improvement on ability to eat (from 12% to 24%, $p=0.02$), to speak (from 25% to 43%, $p=0.007$), the rate of no pain raised from 46% to 57%, the rate of no discomfort raised from 21% to 34% ($p=0.04$) and the score ≤ 5 of OMD raised from 47% to 72% ($p=0.0003$).

Conclusions: The incidence of OMD in Sana'a city is remarkably high, predominantly male, and peaks at 30, 40, and 50 years of age. Jaw deviation dystonia was the dominant type followed by jaw closing dystonia. After the current treatment outcome, the ability to eat has improved, the rate of pain and discomfort has decreased.

Keywords: Oromandibular dystonia (OMD), prevalence, Sana'a city, treatment out-come, Yemen.

INTRODUCTION

A neurological movement disease called dystonia is characterized by recurrent, repetitive, or patterned muscular contractions that cause aberrant movements, postures, or both. The third most common movement

ailment is dystonia, which comes in third place behind essential tremor and Parkinson's disease. Dystonia can be divided into three categories: idiopathic, acquired, or inherited; focal, segmental, multifocal, generalized, and hemi; and age at which symptoms first appear, including infancy, youth, adolescent, early adulthood,

and late adulthood¹. Oromandibular dystonia is a form of focal dystonia that involves the masticatory system, affecting the jaws and/or lingual muscles. Its clinical manifestations may present as lip pursing, tongue dystonia, jaw opening dystonia, jaw closure dystonia, lateral jaw deviation dystonia, jaw protrusion dystonia, or a combination of these abnormal movements². The masseter, temporalis, and to a lesser extent the medial pterygoid muscles are responsible for closing the jaw. The muscles that make up the muscular floor of the oral cavity, the lateral pterygoid and suprahyoid muscles, contract to open the jaw. Pterygoid muscles, especially the lateral ones, are responsible for protrusion and lateral shifts. Most lingual dystonia patients exhibit dystonic contraction in the genioglossus. It affects the ability to chew, speak, and swallow, which causes cosmetic disfigurement, social withdrawal, and negative consequences for the quality of life^{3,4}.

OMD can have a primary (idiopathic) or secondary etiology. The primary form of OMD is the most prevalent type and does not involve any brain lesions, tumors, or pathologies of the central nervous system. OMD's cause and mechanism are not well understood. OMD can develop independently as a neurological ailment (with or without a family history), or it can develop as a side effect of specific medications or conditions like trauma or Wilson's disease⁵. Oromandibular dystonia is estimated to affect 0.1 to 6.9 out of every 100,000 people in western countries^{6,7}. In Arabic countries including Yemen, there have been no data in the incidence of oromandibular dystonia among the Arabic population. Despite prior studies on dental caries⁸, oral and facial abscesses of odontogenic origin^{9,10}, localized aggressive gingivitis (LAP)^{11,12}, periodontitis¹³, bacterial and fungal oral infections¹⁴, interleukin-1 levels in human gingival sulcus¹⁵, and prevalence of signs of temporomandibular disorders in healthy¹⁶ as well, there are no data on the incidence, demographics, clinical characteristics, and treatment outcomes of patients with OMD. Therefore, the purpose of this study was to ascertain the prevalence, demographics, clinical characteristics, and therapeutic outcomes of mandibular dystonia (OMD) patients in Sana'a, Yemen.

SUBJECTS AND METHODS

The Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Sana'a University Clinics, Departments of Oral, Maxillofacial and Neurosurgery at Kuwait University Hospital, and a few private centers for maxillofacial medicine and neurosurgery in the city of Sana'a were all included in the prospective cross-sectional study, which involved 100 Yemeni adults. The study was conducted in 2022.

The study comprised patients with a verified diagnosis of OMD who were routinely followed up at particular clinics. The chosen patients complained of masticatory, lingual, and/or lower facial muscular contraction or involuntary movement. Yemeni patients were included in the investigation.

Data collection: Each patient got a clinical evaluation, and all information was gathered on a case sheet with pooled data that was intended for systematic recording. Age, sex, age at illness beginning, and disease duration were considered demographic factors, whereas OMD type, etiology, family history, personal habits, kind of OMD, prior and current treatments, and their results, afflicted muscles, sensory trick, and task specificity were considered clinical factors. The etiology and kind of dystonia were determined by going over each patient's medical history, neurological examination, and brain imaging results. Additionally, movement disorder specialists used electroneuromyography to identify the affected muscles.

Data analysis: Epi-Info-Version 7 (CDC) was used for the statistical analysis. Count, percentage, mean, and standard deviation (median and minimum, where necessary) were used to sum up categorical measurements to assess the effectiveness of recent treatment and post-OMD care. The percentage difference, 95% confidence interval, and tests for significance using the chi-squared test (X^2) and p -value were used to determine the rates and level of improvement. The cutoff for statistical significance was $p < 0.05$.

Ethical approval: The Medical Ethics Committee of the Faculty of Dentistry at Sana'a University provided its official clearance on November 1, 2021, with the reference number 2021-29. Each study participant signed a consent form. All information, including clinical information and patient identification, was kept private.

RESULTS

The study included 100 cases of OMD diagnosed in one year (2022), out of the estimated population served by the selected clinics, which was, according to the 2014 census, approximately 540,000 people, so the incidence rate of OMD is 100/540,000, equal to 19/100,000/year.

Table 1 shows the age and sex distribution of OMD patients. Most of the patients (92%) are male while only 8% are female. Considering ages, the mean \pm SD for the total patients was 42.4 \pm 10.04 years, for the female group it was 48.1 \pm 8.4 years while the male group had 41.9 \pm 10.1 years less than the female group.

The patient's age ranged from 16 to 70 years. Most patients were in the 40-49 age group (31%), followed by 31-39 years (29%) and 50-59 years (22%), while OMD was less frequent in the younger or older age groups. Table 2 shows the socio-demographic data of OMD patients attending the Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Sana'a University Clinics and Departments of Oral, Maxillofacial and Neurosurgery at Kuwait University Hospital and some private centers for maxillofacial medicine and neurosurgery in the city of Sana'a during the year of 2022. The proportion of males is 92% and females 8%, and most of the patients had university education (33%), secondary education (37%) and 26% primary education.

Table 1: Age distribution of the OMD patients.

Age groups	Male n=92	Female n=8	Total
	N (%)	N (%)	N (%)
16-30 years	12 (13)	0 (0)	12 (12)
31-39 years	29 (31.5)	0 (0)	29 (29)
40-49 years	27 (29.3)	4 (50)	31 (31)
50-59	20 (21.7)	2 (25)	22 (22)
≥60	4 (4.3)	2 (25)	6 (6)
Total	92 (92)	8 (8)	100 (100)
Mean	41.9 years	48.1 years	42.4 Years
SD	10.1 years	8.4 years	10.04 years
Median	40 years	47.5 years	40 years
Mode	40 years	40 years	40 years
Min-Max	16-70 years	40-60 years	16-70 years

Table 2: Socio-demographic data of OMD patients.

Characteristics		Frequency (%)
Gender	Males	92 (92)
	Females	8 (8)
Education level	No education	4 (4)
	Primary school	26 (26)
	Secondary school	37 (37)
	University	33 (33)
Marital status	Married	97 (97)
	Single	3 (3)
	Divorced	0 (0)
Current employment	Employment	78 (78)
	Retired	0 (0)
	Unemployment	22 (22)

Most OMD patients were married (97%), Single patients only 3%, and no divorced case. Looking at current employment, 78% were employed and only 22% were unemployed.

Table 3 shows the social habits and oral habits of OMD patients attending the selected maxillofacial surgery, dental clinics during a year. Qat chewing was prevalent as 95% of OMD patients had this habit, 56% for 5 hours or less per day, 30% for 6-10 hours per day and 9% for 11 or more hours per day (hours chewing time/day). Considering the smoking habit, 47% of OMD patients are smokers, 80.9% are light smokers and 19.1% are heavy smokers. When considering oral habits 19% of OMD patients had bruxism and 4% clenching.

Table 3: Social habits and oral habits of OMD patients attending maxillofacial, Faculty of Dentistry, Sana'a University Clinics and some maxillofacial centers in Sana'a city during a year.

Characteristics		Frequency (%)	
Qat chewing	Yes	95	95
	No	5	5
Frequency per day	One	74	74
	Two	21	21
Time of chewing hours/day	≤ 5 hours	56	56
	6-10 hours	30	30
	≥11 hours	9	9
	Mean time of chewing=5.9 hours, Min=2 hours, Max=20 hours		
Smoking	Light –moderate <20/day	38	80.9
	Heavy >20 /day	9	19.1
	Total	47	47
Shamah	Yes	10	10
	No	90	90
Oral Habits	No	48	48
	Bruxism	19	19
	Clenching	4	4
	Bruxism+ Clenching	29	29

Table 4 shows family history of OMD, onset and modes of onset of OMD among our patients. Only 7% of OMD patients have a family history of OMD. Looking at the first time MOD onset, 33% of patients had it before 12 months or less, 21% before 13-24 months, while the majority (46%) had MOD before ≥25 months; with mean±SD=50.9±64.9 months, and ranged from 2 months to 420 months (35 years). In 4% of OMD patients the first episode onset is after an accident, 29% after stress, 11% after illness, 6% after dental treatment, and 48% without causes. After the

onset of MOD, 94% of patients were seeking treatment, while 6% were not. Looking at previous and subsequent treatments, most patients were treated with medication (86%), 47% tried Botox, 18% tried acupuncture, 51% tried physical therapy and 8% underwent surgical treatment. When considering treatment with Botox, no improvement was seen in 55.3% of MOD patients who used it, 19.1% showed slight improvement, 21.3% showed moderate improvement, and 4.2% showed significant improvement. When considering drug therapy, 53.5%

of MOD patients showed no improvement, 19.8% showed little improvement, 19.8% showed moderate improvement, and 8.1% showed significant improvement. When considering previous acupuncture

treatment, 72.2% of MOD patients who used it showed no improvement, 22.2% showed slight improvement, 5.6% showed moderate improvement while none (0%) showed significant improvement.

Table 4: Family history, first time occurrence and methods of appearance of OMD among all patients.

Characters		Number (%)
Family History	Yes	7 (7)
	No	93 (93)
First time onset (before) Months	≤12 months	33 (33)
	13-24 months	21 (21)
	≥25 months	46 (46)
Onset Mean ± SD= 50.9±64.9 months, Min=2 months, Max=420 months		
How OMD occurrence (Type of onset)	After Accident	4 (4)
	*Head trauma	2 (2)
	*Blow in lower jaw	2 (2)
	After surgery	0 (0)
	After illness	11 (11)
	*Ischemic stroke	1 (1)
	*Psychiatric disorder	4 (4)
	*Temporomandibular disorder	1 (1)
	*Sever dental pain	2 (2)
	*Hypothermia " exposure to cold or wind"	3 (3)
	After dental treatment	6 (6)
	*Oral surgery	1 (1)
	*Dental extraction	2 (2)
	*Endodontic treatment	1 (1)
	*Fixed prosthesis	3 (3)
	After heavy Qat chewing	2 (2)
	After stress	29 (29)
No reason	48 (48)	
Seeking Treatment	Yes	94 (94)
	No	6 (6)
Type of treatments	Botox	47 (47)
	MAB	0 (0)
	Medication	86 (86)
	Acupuncture	18 (18)
	Physiotherapy	51 (51)
	Surgical therapy	8 (8)

When considering physiotherapy, no improvement was seen in 64.7% of MOD patients, 27.5% showed slight improvement, 4% showed moderate improvement, and 4% showed significant improvement. When considering surgical treatment, 75% of MOD patients showed no improvement, 0% showed little improvement, 12.5% showed moderate improvement, and 12.5% showed significant improvement.

When considering treatment with oral appliance of pphysiotherapy, no improvement was seen in 62.5% of MOD patients who used it, 29.2% showed slight improvement, 4.1% showed moderate improvement, and 4.1% showed significant improvement. When considering massage, 53.3% of MOD patients showed no improvement, 33.3% showed little improvement, 13.3% showed moderate improvement, and 0% showed significant improvement. When considering Physical exercise, 50% of MOD patients who used it showed no improvement, 50% showed slight improvement, and no moderate improvement or significant improvement seen (Table 6). When taking into account the type of medication anticholinergics and baclofen were the most often used drugs, followed by anticonvulsants at 27.9%, benzodiazepines at 26.7%, and vitamins at 22.1% [patients who utilized medication (86%)].

However, among all participants, the use of muscle relaxants, tricyclic antidepressants, steroidal analgesics, non-steroidal analgesics, and dopaminergics ranged from 1 to 19% (Table 7).

From the total number of patients, jaw deviation dystonia accounted for 65% of cases, followed by jaw closing dystonia (42%), jaw protrusion dystonia (29%), and jaw opening dystonia (27%), while tongue dystonia and lip dystonia were less common (16% and 11%, respectively). When multiple dystonias were taken into account, 47% of patients had more than one type. The most common type was "jaw closing dystonia+ jaw deviation dystonia," which accounted for 26% of cases. Other common types included "jaw closing dystonia + tongue" (5%), "jaw closing dystonia +jaw protrusion dystonia," "jaw closing dystonia+ jaw deviation dystonia + jaw protrusion dystonia," and " jaw closing dystonia + jaw deviation dystonia + jaw protrusion dystonia" (1%), were rare (Table 8).

Considering assessment the mastication as rating scale used to comprehensively assess for OMD before starting current treatment and after current treatment. There was improvement on ability to eat but slow from 12% to 24% after treatment in which difference %

was 12, with 95% CI ranged from 4.9-27, and the difference was significant ($X^2=4.9$, $p=0.02$). Also there was decrease in patients ability to eat soft food but slow from 58% prior treatment to 28% after

treatment in which difference % was 28, with 95% CI ranged from 14.2-40.2, and the difference was highly significant ($X^2=15$, $p=0.0001$).

Table 5: Past and present seeking of treatments and type of treatment used by OMD patients and responses, (n=100).

		No. (%)
Seeking Treatment	Yes	94 (94)
	No	6 (6)
Type of past and present treatment	Botox	47 (47)
	No improvement	26 (55.3)
	Mild improvement	9 (19.1)
	Moderate improvement	10 (21.3)
	Marked improvement	2 (4.2)
	MAB	0
	No improvement	0
	Mild improvement	0
	Moderate improvement	0
	Marked improvement	0
	Medication	86 (86)
	No improvement	45 (53.5)
	Mild improvement	17 (19.8)
	Moderate improvement	17 (19.8)
	Marked improvement	7 (8.1)
	Acupuncture	18 (18)
	No improvement	13 (72.2)
	Mild improvement	4 (22.2)
	Moderate improvement	1 (5.6)
	Marked improvement	0
	Physiotherapy	51 (51)
	No improvement	33 (64.7)
	Mild improvement	14 (27.5)
	Moderate improvement	2 (4)
	Marked improvement	2 (4)
	Surgical therapy	8 (8)
	No improvement	6 (75)
Mild improvement	0	
Moderate improvement	1 (12.5)	
Marked improvement	1 (12.5)	
Type of surgery used	Maxillomandibular fixation (MMF)	12.5
	Arthrocentesis	25
	TMJ surgery	25
	Bilateral coronoidectomy	25
	TMJ surgery+ Bilateral coronoidectomy	12.5

Table 6: Physiotherapy treatments and used by OMD patients and responses, (n=51).

Physiotherapy	Response	No. (%)
Oral appliance		51(51)
	No improvement	24 (47.1)
	Mild improvement	15 (62.5)
	Moderate improvement	7 (29.2)
	Marked improvement	1 (4.1)
Massage		30 (58.8)
	No improvement	16 (53.3)
	Mild improvement	10 (33.3)
	Moderate improvement	4 (13.3)
	Marked improvement	0
Physical exercise		6 (11.8)
	No improvement	3 (50)
	Mild improvement	3 (50)
	Moderate improvement	0
	Marked improvement	0

Table 7: Past and present medication of treatments and type of medications used by OMD patients and responses, (n=100).

	No. (%)
Response of past medication used	86 (86)
No improvement	45 (53.5)
Mild improvement	17 (19.8)
Moderate improvement	17 (19.8)
Marked improvement	7 (8.1)
Type of medications used	
Anti-cholinergic	42 (48.8)
Baclofen	42 (48.8)
Anticonvulsant	24 (27.9)
Benzodiazpine	23 (26.7)
Vitamins	19 (22.1)
Tricyclic antidepressant	17 (19.7)
Muscle relaxant	13 (15.1)
Analgesic-non-steroidal	10 (11.6)
Anti- Dopaminergic	7 (8.1)
Analgesic-steroidal	5 (5.8)
Dopaminergic	1 (1.2)

Table 8: Prevalence the different type of dystonia among the current study patients, (n=100).

Types	No. (%)
	86(86)
Jaw deviation dystonia	65 (65)
Jaw closing dystonia	42 (42)
Jaw protrusion dystonia	29 (29)
Jaw opening dystonia	27 (27)
Tongue dystonia	16 (16)
Lip dystonia	11 (11)
Multi-dystonia	47 (47)
Jaw closing dystonia+ Jaw deviation dystonia	26 (26)
Jaw closing dystonia + Tongue	5 (5)
Jaw closing dystonia +Jaw protrusion dystonia	5 (5)
Jaw closing dystonia+Jaw deviation dystonia+ Jaw protrusion dystonia	4 (4)
Jaw closing dystonia + Lip dystonia	3 (3)
Jaw closing dystonia+ Jaw deviation dystonia + Tongue dystonia	2 (2)
Jaw closing dystonia+ Jaw opening dystonia	1 (1)
Jaw closing dystonia+ Jaw deviation dystonia + Lip dystonia	1 (1)

Finlay 6% of patients change to normal mastication scale with, difference % equal to 6, 95% CI ranged from 1.1-12.4, and the difference was significant ($X^2=6.1$, $p=0.013$). When considering speech assessment as a rating scale used to comprehensively assess OMD before the start of current treatment and after current treatment to confirm improvement after treatment. The rate of normal speech rose from 25% to 43% after treatment with, difference % equal to 18, 95% CI ranged from 4.8–30.3, and the difference was highly significant ($X^2=7.1$, $p=0.007$). There was an improvement in the ability to speak among the patients as the rate of audible but difficult comprehend decreased from 25% before treatment to 10% after treatment where difference was 15%, with 95% CI ranged between 4.5-25.3, and the difference was highly significant ($X^2=7.7$, $p=0.005$). Also, there was an improvement in the ability to speak among the patients as the rate of Inaudible (less 50%) decreased from 10% before treatment to 2% after treatment where difference was 8%, with 95% CI ranged between 1.2 – 15.6, and the difference was significant ($X^2=5.6$, $p=0.017$). When considering pain assessment as a rating scale used to comprehensively assess OMD before the start of current treatment and after current treatment to confirm improvement after treatment. The rate of no

pain raised from 46% to 57% after treatment with, difference % equal to 11%, 95% CI ranged from -2.8 – 24.2, and the difference was not statically significant ($X^2=2.4$, $p=0.12$). However, there was an improvement in the rate of moderate, intermittent to continuous pain in which it significantly decreased from 12% prior to treatment to 3% after treatment where difference was 9%, with 95% CI ranged between 1.6-17.1, and the difference was significant ($X^2=5.8$, $p=0.015$). Also, there was an improvement in the severe pain as the rate decreased from 3% before treatment to 1% after treatment but result was not statically significant ($X^2=1.01$, $p=0.31$)

When considering OMD assessment as a rating scale used to comprehensively assess OMD before the start of current treatment and after current treatment to confirm improvement after treatment. The rate of no discomfort raised from 21% to 34% after treatment with, difference % equal to 13%, 95% CI ranged from -1.6-24.8, and the difference was statically significant ($X^2=4.2$, $p=0.04$). Also, there was an improvement in the rate of ‘ Moderate to severe discomfort’ in which it significantly decreased from 24% prior to treatment to 9% after treatment where difference was 15%, with 95% CI ranged between 4.7-25.1, and the difference was highly significant ($X^2=8.1$,

$p=0.004$). Also, there was an improvement in the severe discomfort as the rate decreased from 11% before treatment to 1% after treatment and the result was statically significant ($X^2=8.8$, $p=0.003$). When considering OMD assessment as a score scale used to comprehensively assess OMD before the start of current treatment and after current treatment to confirm improvement after treatment. The mean \pm SD of discomfort score was 6.4 ± 2.9 score before treatment decreased to 4.1 ± 2.7 scores after treatment, indicating improvement. The score ≤ 5 of discomfort score raised from 47% to 72% after treatment with,

difference % equal to 25%, 95% CI ranged from 11.4 – 37.3, and the difference was statically significant ($X^2=12.9$, $p=0.0003$), the 6-10 score of discomfort decreased from 41% to 23% after treatment with, difference % equal to 18%, 95% CI ranged from 5.1 – 30.1, and the difference was statically significant ($X^2=7.4$, $p=0.006$). The ≥ 11 score of discomfort decreased from 12% to 5% after treatment with, difference % equal to 7%, 95% CI ranged from -0.94– 15.3, and the difference was not statically significant ($X^2=3.1$, $p=0.07$).

Table 9: Rating scale used to comprehensively assess OMD before starting current treatment and after current treatment, (n= 100).

Scales	Before N (%)	After N (%)	Difference %	95% CI	X ²	p	
Mastication	Normal	0 (0)	6 (6)	6	1.1-12.4	6.1	0.013
	Able to eat but slow	12 (12)	24 (24)	12	4.9-0.027	4.9	0.02
	Able eat soft food	31(31)	23 (23)	8	-4.3-19	1.6	0.2
	Able eat soft food but slow	58 (58)	30 (30)	28	14.2-40.2	15	0.0001
	Only able consume liquid	0 (0)	0 (0)	0	-3.7-3.7	0	1.0
Speech	Normal	25	43(43)	18	4.8-30.3	7.1	0.007
	Find hard speak clear	37 (37)	27 (27)	10	-2.9-22.4	2.3	0.13
	Audible but difficult comprehend	25 (25)	10 (10)	15	4.5-25.3	7.7	0.005
	Inaudible (less 50%)	10 (10)	2 (2)	8	1.2-15.6	5.6	0.017
	Inaudible (more 50%)	3 (3)	1 (1)	2	-2.8-7.5	1.01	0.31
Pain	No pain	46 (46)	57 (57)	11	-2.8-24.2	2.4	0.12
	Mild intermittent	26 (26)	17(17)	9	-2.4-20.1	2.4	0.12
	Mild continuous to moderate	14 (14)	6 (6)	8	-0.48-16.7	3.5	0.06
	Moderate, intermittent to continuous	12 (12)	3 (3)	9	1.6-17.1	5.8	0.015
	Severe	3 (3)	1 (1)	2	-2.8-7.5	1.011	0.31
Discomfort	No Discomfort	21 (21)	34 (34)	13	1.6-24.8	4.2	0.04
	Mild	19 (19)	24(24)	5	-6.4-16.7	0.73	0.39
	Mild to moderate	27 (27)	16 (16)	11	-0.4-22.1	3.6	0.059
	Moderate to severe	24 (24)	9 (9)	15	4.7-25.1	8.1	0.004
	Severe	11 (11)	1 (1)	10	3.5-17.7	8.8	0.003
OMD scores	≤ 5 score	47 (47)	72 (72)	25	11.4-37.3	12.9	0.0003
	6-10 score	41 (41)	23 (23)	18	5.1-30.1	7.4	0.006
	≥ 11 score	12 (12)	5 (5)	7	-0.94-15.3	3.1	0.07

Before: Mean \pm SD= 6.4 ± 2.9 score, Min=2 score, Max=13 score

After: Mean \pm SD= 4.1 ± 2.7 score, Min=0 score, Max=13 score

Electromyography (EMG) often shows excessive muscle activity, but it is not conducted in our study because the findings are not specific to dystonia. Current treatment All types of treatment modalities except MAB, and our team acted as a therapist and monitor.

DISCUSSION

OMD is a rare disease, which decreases quality of life by affecting eating, drinking, speaking and swallowing functions^{17,18}. In this study, the clinical and demographic data of 100 patients with OMD who were followed up at our outpatient clinics in Sana'a city were determined. To the best of our knowledge and after searching in medical data bases, this is the first study in Yemen and this is one the largest cohort after those in the studies of Balal *et al.*, Scorr *et al.*, Slaim *et al.*, Yoshida *et al.*, and Tan *et al.*,^{17,19-22}. In the current study, the incidence of OMD is 19/100,000/year; this incidence is higher than that reported elsewhere as in Turkey in which the annual

incidence was reported to be between 3.3 and 6.9 per million people^{17,23}. Most of our patients (92%) were males while only 8% were females; this result is different from that reported with the literature^{19,22,24}. The high prevalence of oromandibular dystonia is strongly correlated with the chewing of Qat, and patients' reluctance to give up the habit prevents them from getting better and may make their condition worse. Also, cases that typically stop consuming khat, feel better, and react to treatment. Rarely, they improve with khat use and deteriorate when it is stopped. In the current study, the mean \pm SD for the total patients was 42.4 ± 10.04 years, and most patients were in the 40-49 age group (31%), followed by 31-39 years (29%) and 50-59 years (22%) which indicate that in our

patients the onset of the disease was commonly in 4th decade and in the 3rd and less number in 5th decade, this finding is in contrast with that reported by Slaim *et al.*,¹⁹, Tan and Jankovic²⁰ Scorr *et al.*,²¹ and Balal *et al.*,¹⁷ in which the onset of the disease was reported to be common after the 5th decade. The lower mean age in our patients than that previously reported can be explained by the younger age of post-anoxic, post-traumatic and neurodegenerative patients. Moreover, men were younger than women [mean age for women was 48.1±8.4 years older than the male mean (41.9±10.1 years)], this may be because secondary etiologies were more common in men as qat chewing, stress than in women and this explain why OMD is more common among men in Yemen.

When looking at the different types of dystonia from the total number of patients, jaw deviation dystonia was dominant at 65%, Our result is in contrast with the literature, in which the most common type of OMD was JO dystonia^{17,19-22,26,27}. This was followed by followed by jaw closing dystonia (42%), jaw protrusion dystonia (29%), and jaw opening dystonia (27%). Our data are consistent roughly with those of Sinclair *et al.*,²¹. The prevalence of multi-dystonia was high among our patients of OMD (47%), particularly Jaw closing dystonia + Jaw deviation dystonia (26%). The data were consistent with those in the literature^{19,24,27,28}. In addition, the association between two types of dystonia or more caused a significant decrease in the quality of life of patients¹⁷. Chewing, swallowing, and speech are frequently impaired in OMD, altering quality of life and social relationships^{19,28}. In current study patients, 75% of patients had speech impairment, 100% had mastication disorders, 79% suffering from pain, and 81% suffering from discomfort (Table 9). Our results are different from that reported by Salim *et al.*,¹⁹ in which low rate of mastication disorders and pain (32.5%). Also Sinclair *et al.*, described different results with speech and eating impairment in 54.3% of patients and pain in 34.3%²⁶. The causes of dystonia can also be fairly diverse. The majority are idiopathic, meaning that despite rigorous diagnostic testing, no cause could be identified. There are numerous probable reasons, both acquired and hereditary, even if a cause can only be identified in a small percentage of cases^{17,19}. Half of the patients in our group (48%) presented as idiopathic OMD and the other half had etiologies listed in Table 4. The secondary form was the most common and it was after stress (29%). Tan and Jankovic²⁴ and Sliam *et al.*,¹⁹ noticed different results with 63.0% and 73% of patients having idiopathic OMD. Physical exercise was performed for 6 patients, 50% of whom used it showed no improvement, 50% showed slight improvement, and no moderate or significant improvement was observed (Table 6). Patients with dystonia often request physical therapy. These actions seem very intuitive in view of the spasms and pain caused by excessive involuntary muscle contractions. Frequently requested options include strengthening of non- dystonic muscles, "hold" exercises, and stretching of tight muscles. Several small open trials of various physical therapy modalities have reported positive results for different types of

dystonia, but more rigorous studies have failed to demonstrate any lasting benefits^{29,30}. Although there is no evidence, it is reasonable to offer physiotherapy according to the patient's specific needs, especially those with severe generalized dystonia who may develop contractures³¹.

In the current study, when considering previous and subsequent acupuncture treatment, 72.2% of MOD patients who used it showed no improvement, 22.2% showed slight improvement, 5.6% showed moderate improvement. Also when considering previous and subsequent physiotherapy, no improvement was seen in 64.7% of MOD patients, 27.5% showed slight improvement, 4% showed moderate improvement, and 4% showed significant improvement (Table 5). Patients frequently inquire about associated practices as well, including chiropractic care, yoga, meditation, and other programs for stress relief or relaxation³². Even while some patients do prefer these options, there is little evidence of a long-term benefit in this case. With the exception of chiropractic techniques, which can occasionally be quite harsh or unpleasant, there is typically no danger in these operations. When considering previous and subsequent drug therapy (medications, 86 patients gone through courses of medications) 53.5% of MOD patients showed no improvement, 19.8% showed little improvement, 19.8% showed moderate improvement, and 8.1% showed significant improvement (Table 5). Numerous drugs are available for dystonia patients^{31,33-40}. Despite the fact that evidence-based evaluations have been published, no existing agent has undergone thoroughly controlled clinical testing. Their use is primarily determined by anecdotal evidence and professional judgment. There are two major groups into which the therapeutic alternatives might be divided. These include symptomatic medications that are helpful for many different types of dystonia, such as oral benzodiazepines, muscle relaxants, antispasticity agents, anticholinergics, and levodopa³¹, as well as medications that are only beneficial in certain situations, such as inherited dystonias, for which specific treatments include removing a toxic substance, taking vitamins and/or cofactors, and avoiding triggers medications³¹. Anticholinergics and baclofen were the most often prescribed drugs in our study (48.8% each), followed by anticonvulsants (27.9%), benzodiazepines (26.7%), and vitamins (22.1%) (Table 7). There was significant improvement on each of the markers of the rating scale used to evaluate OMD in its whole before and after current treatment, including mastication, speech, pain, and discomfort (Table 9). All dystonias are curable, according to the overall conclusion drawn from our Table 9 results. Most patients benefit at least in part from symptom-based care, and certain uncommon subtypes have unique therapies that target the underlying pathophysiologies of the disease. The range of treatment options is anticipated to expand in the near future for both adults and children as continuing research enhances our capacity to exploit current medicines and identifies new causes and biological systems where new interventions can be developed⁴¹.

Limitation of the study

A limitation of this study was that the patients' actual medical history, medications, and habits were determined by their question and there is a possibility that they may not be telling the truth or have hidden information. It was also difficult to conduct laboratory tests for all cases to confirm the information received because of the type of study (cross-sectional study), and because of the large sample size. Also, not all patients regularly have medical cheek exams, and some people don't even know about their medical health.

CONCLUSIONS

The incidence of OMD in Sana'a city is remarkably high, predominantly male, and peaks at 30, 40, and 50 years of age. Jaw deviation dystonia was the dominant type followed by jaw closing dystonia. After the current treatment, the ability to eat and speak have improved the rate of pain and discomfort has decreased. This study adds more data to the literature by identifying the clinical features of this rare disorder in Yemen and draws attention to this neglected type of dystonia. It is important to treat the underlying cause of the disorder, for example, in cases of malocclusion due to a fixed prosthesis. There was a remarkable response to medication after correction of the obstruction. The patient runs the risk of getting sick again, even after a full recovery. The cause may be related to psychological or environmental factors, such as exposed to cold. Since the illness is brought on by involuntary movements connected to the central nervous system, systemic and local treatment is preferable rather than local treatment only. The better the reaction to treatment, the lower the rating scale of symptoms. It is preferable to use more than one method of treatment at the same time rather than one treatment in separate periods.

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AUTHOR'S CONTRIBUTIONS

Sharaf-Aldeen HMA: clinical work and followed up with patients as part of a master's degree at Sana'a University in Yemen. **Abbas AKMA:** acquired the data. **Al-Kibsi TAM:** formal analysis, editing. **Al-Shamahy HA:** investigation, conceptualization. **Jahaf SHA:** analyzed the data and interpreted the results. **AL-Kaff RHSO:** data curation, investigation. All authors revised the article and approved the final version.

DATA AVAILABILITY

The data and material are available from the corresponding author on reasonable request.

CONFLICT OF INTEREST

None to declare.

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