Temporomandibular Disorder Related Characteristics and Treatment Outcomes in Oromandibular Dystonia Patients in Two Different Clinical Settings: A Cross-Sectional Study

A Thesis Submitted to the Faculty

Of the University of Minnesota by

Asha Balu Sude

In partial fulfillment of the requirements for the degree of

Master of Science

Donald R Nixdorf

April 2020

Acknowledgements

I am deeply grateful to Dr. Donald Nixdorf who has provided guidance and invaluable inspiration for my M. S. thesis. This project would not have been possible without his support. My sincere gratitude to my thesis committee members, Dr. Shanti Kaimal and Dr. Ashley Petersen, for their valuable time, insights and suggestions on this project.

Special thanks to Dr. Joseph Matsumoto for his expertise and tremendously helpful collaboration with Movement Disorder clinic. I am deeply indebted to Dr. Eric Schiffman, Dr. Estephan Moana-Filho, Dr. Cory Herman, and Dr. Mariona Mulet for providing critical reviews that enhanced the clarity of this project. I would also like to thank my amazing co-residents and wonderful staff of Orofacial Pain and Movement Disorder clinic at University of Minnesota for their immense support.

Abstract

Introduction: Clinical presentation of oromandibular dystonia is variable and it can be further complicated with presence of TMD symptoms. We sought to evaluate variations in clinical presentation of OMD patients, particularly TMD related characteristics, in two clinic settings.

Methods: In a cross-sectional study design, a web-based data collection survey was provided to eligible patients with OMD from Movement Disorder and Orofacial Pain clinics. Patients with OMD were identified using ICD-9 and ICD-10 diagnostic codes from their electronic health record system. We excluded patients <18 years old, pregnant women, prisoners, non-English speakers, patients with dementia, OMD diagnosis secondary to Parkinson's disease and Tardive dyskinesia patients. The survey questionnaire was designed to collect information on demographic characteristics, clinical presentation particularly related to TMD, quality of life and treatment outcomes. Validated questionnaires were used when available such as TMD Screener, EQ-5D-5L, jaw functional limitation scale, and global rating of change scale.

Results: Of 53 eligible patients; 31 responded to the survey for a 59% response rate. 48% patients in MD clinic and 60% patients in OFP clinic reported experiencing jaw pain along with involuntary movements. Of those, 90% from MD group and 83% from OFP group screened positive with TMD pain screener. TMD screener remained positive in about 40% patients in both clinics in the last 30 days of questionnaire response. Global score of JFLS was higher in both groups. None of the study comparisons between two groups were statistically significant.

Conclusion: Patients with OMD have features of TMD, irrespective of the clinical setting they seek and receive care from. OMD patients from both clinics were similar in terms of clinical presentation, quality of life and treatment outcomes.

Table of Contents

	Page
List of Tables	v
List of Abbreviations	vi
Introduction	1
Methods	2
Results	7
Discussion	11
Conclusion	15
Flow chart	16
List of Tables	17
Bibliography	31

List of Tables

		Page
Table 1	Demographic questionnaires	17
Table 2	General Health Status	18
Table 3	EQ-5D-5L Summary score	19
Table 4	Symptom description before getting treatment	20
Table 5	Clinical Care	24
Table 6	Symptoms after getting treatment	26
Table 7	TMD screener 1	28
Table 8	TMD screener 2	28
Table 9A	TMD related questions	29
Table 9B	Global score of Jaw Functional Limitation Scale	29
Table 9C	Oral Behavioral Checklist	30

List of Abbreviations

OMD	Oromandibular dystonia
TMD	Temporomandibular disorders
MD	Movement disorder
OFP	Orofacial pain
DC/TMD	Diagnostic criteria for Temporomandibular disorders
JFLS	Jaw Functional Limitation Scale
QoL	Quality of Life
EQ-5D-5L	EuroQol 5-Dimensions 5-Levels
GBI	Glasgow Benefit Inventory
OMDQ-25	Oromandibular dystonia questionnaire-25
CR-QoL	Communication-Related Quality of Life
TSV	Tab-separated values
JASP	Jeffery's Amazing Statistics Program

INTRODUCTION

Oromandibular dystonia (OMD) involves sustained or repetitive involuntary movements of the masticatory, facial, and lingual muscles. Different population studies of OMD have shown prevalence of 2.1 to 6.9 per 100,000 persons (1-3). Clinical characteristics of OMD are a reflection of different muscles undergoing dystonic spasm causing involuntary movements like opening, closing, side or protrusive jaw movements, facial grimacing, tongue movements, lip movements like pursing, sucking or smacking, and retraction of corners of mouth (4). Data regarding occurrence of TMD or jaw pain in OMD patients is variable in literature (5-7). TMD was often considered in differential diagnosis of OMD, particularly to jaw opening dystonia due to similarity with temporomandibular joint subluxation (4). Costa and colleagues found that patients with oromandibular dystonia frequently report some form of TMD (8). Given the increasing reports of occurrence of TMD in OMD patients and heterogeneity in clinical characteristics of OMD, we intend to study variation in clinical presentation of OMD in Movement Disorder (MD) clinic and OFP clinic.

Involuntary movements in OMD typically appear and worsen with jaw functions that impart negative effects on quality of life (9, 10). Despite the

significant disease burden, there is paucity of literature on health status and quality of life of OMD patients (11-13). Since OMD is not curable; treatment is often focused on reducing dystonic movements and improving one's functions. Commonly reported treatment options include botulinum toxin injections, oral medications, oral appliances, physical therapy, behavioral therapy and occasionally deep brain stimulation (14, 15). Therefore, we wanted to explore clinical characteristics, evaluate quality of life and assess treatment outcomes in OMD patients who presented to MD and OFP clinic.

METHODS

This is a cross-sectional survey study where 43 questions about OMD and TMD were asked of eligible participants receiving care within two different services, a Movement Disorder (MD) clinic where care is provided by physicians and an Orofacial Pain (OFP) clinic where care is provided by dentists with advanced training in OFP. Both clinics are part of the University of Minnesota system and located on the East Bank of the Twin Cities campus.

Patient selection

After the approval of institutional review boards, patients with OMD were identified using ICD-9 (333.82 Orofacial dyskinesia) and ICD-10 (G24.4 Idiopathic orofacial dystonia) diagnostic codes from electronic health record systems of MD clinic and OFP Clinic when they were seen between October 2012 to February 2019. Exclusion criteria were patients <18 years old, pregnant women, prisoners, non-English speakers, patients with dementia, OMD diagnosis secondary to Parkinson's disease, Tardive dyskinesia, or other movement disorders like chorea, hemifacial spasm, cervical dystonia, tremor, isolated blepharospasm, and functional movement disorder. We identified 42 patients from MD clinic and 11 patients from OFP clinic who met eligibility criteria (Fig. 1).

Recruitment strategy

Eligible patients were contacted through a recruitment letter for voluntary participation in the survey for data collection. Recruitment letters included information about the research, reasons for conducting the study, benefits of taking part in the survey and our contact details for further questions. The letters had a URL link and QR code for participants to access the web-based

questionnaire. The survey was designed and administered through Qualtrics^{XM}. The survey reminders were sent three additional times at 4-week intervals. Upon completion of the survey, participants were sent a thank you letter along with a $25 gift card to compensate for their time.

Data collection instruments

A 43-items questionnaire was designed to collect information on demographic features (age, gender, ethnicity, education), clinical features of involuntary movements (onset, trigger, kind of involuntary movements, its effect on function, sensory tricks, associated muscular tension, and jaw pain with relation to involuntary movements), clinical care received (number of providers seen, kind of treatments received, most effective treatment, reason for discontinuing treatment), and TMD related characteristics. The questionnaire was divided into categories like symptoms pre-treatment, clinical care received, symptoms post-treatment, general health status and demographics and were based on existing validated questionnaires when available.

The specific instruments used were:

- TMD pain screener (16), which is a 6-item instrument for identifying patients with painful TMD. Scores ranging from 0-7 with threshold value for positive score being 3 or more. TMD pain screener was displayed twice to participants; first to participants when they were experiencing jaw pain along with involuntary movements. Second, when all the participants in both groups were requested to fill out the TMD Screener for the symptoms in the last 30 days irrespective of the onset of jaw pain with involuntary movements.

- Characteristic Pain Intensity from Graded chronic pain scale (17), and a 30-days version from DC/TMD (18) was used for assessment of average pain severity that ranges from 0 (no pain) to 10 (pain as bad as could be). Score ranges from 0 to 100 points.

- Jaw functional Limitation Scale (JFLS) involving 8-item instrument (19) (short version) was used to assess functional limitation of the masticatory system. For each item, the participants rate their limitation on a 0 – 10 point scale, where "0" being no limitation and "10" being severe limitation. A "not-applicable" option was available to participants to indicate that the item is irrelevant for their situation,

which was considered as missing. Global score was computed from the mean of available items with no more than 2-items missing.

- Modified short version of Oral behavioral checklist (20), an instrument for assessing self-reported awareness of the presence and extent of 15 waking state oral behaviors. These behaviors were coded on 0 (none of the time) – 2 (most or all of the time). These scores can range from 0 to 30.

- EQ-5D-5L (21) is a measure of health status developed by EuroQol group that provides a generic measure of clinical and economic burden of disease. It includes 5 dimensions (mobility, self-care, usual activities, pain/discomfort, and anxiety/depression) with 5 levels in each dimension (no problems, slight problems, moderate problems, severe problems, and extreme problems). EQ-5D-5L states will be converted to a summary number or index value using the United States value sets (22). The index value represents how good or bad a health state is according to preferences of the general population in that country (in our case, USA) and it can range from -0.573 (worse than dead) to 1 (perfectly healthy).

- Global rating of change scale (23) involves a single question that measures self-perceived change in health status. It is a 11-point scale ranging from -5 (very much worse), through 0 (unchanged), to +5 (completely recovered).

Data management and analysis

The survey data collected in Qualtrics^{XM} application was downloaded in TSV file format. The collected data was analyzed using JASP statistical software. Descriptive statistics were presented as mean and standard deviation for numerical data and counts and proportions for categorical data. Mann-Whitney's U test was used to compare means due to non-normal distribution of data. Chi-square test was used to compare proportions of two groups.

RESULTS

Demographic features and general health status:

10/11 (91%) patients in OFP clinic and 21/42 (50%) patients in MD clinic responded to our survey with no missing values to any questions for all completed questionnaires. All the participants identified themselves as Caucasians with predominance of females in both groups (3:1 female: male). EQ-5D-5L scale showed no significant difference in summary index score of participants from both groups (p-value 0.374). Both the groups had comparable baseline characteristics and general health status (Tables 1, 2, & 3).

Symptoms before getting treatment:

Survey participants in MD group had noted involuntary movements for longer duration (mean 15 years) compared to OFP group (8 years) (p-value 0.330). Jaw closing and jaw opening movements were more commonly reported in MD group while jaw closing, tongue movements were often reported in OFP group. On an ordinal scale, involuntary movements bothered "a lot" to 60% in OFP group compared to 43% in MD group. (p-value 0.313). 67% participants in the MD group had sensory tricks compared to 50% participants in OFP (p-value 0.373) (Table 4).

Muscle tension associated with involuntary movements was reported by 71%

participants in MD group compared to 90% participants in OFP group (p-value 0.248). However, pain associated with involuntary movements was reported by 48% (10 out of 21) participants in MD group compared to 60% (6 out of 10) in OFP group (p-value 0.519). These 16 participants who experienced pain with involuntary movements were further asked six-item TMD screening instrument. In the MD group, 9 of 10 screened positive for TMD with threshold value of 3 or more as opposed to 5 of 6 in OFP group (p-value 0.696). Involuntary movements bothered participants more than jaw pain in 7 out of 10 from MD group and 3 out of 6 in OFP group. Further, involuntary movements were the chief reason for seeking healthcare in 8/10 participants in MD group and 3/6 participants in OFP group (Tables 4 & 7).

Clinical Care:

The average number of providers seen by participants in MD group was 8 (range 1-25) compared to 5 (range 3-7) in OFP group (p-value 0.231). Botulinum toxin injection was the most commonly received treatment in both groups followed by medications in MD group and mouthguards in OFP group. Botulinum toxin injections were the most effective treatment per participant self-report (MD-48%, OFP-40%, p-value 0.350). Up to 76% of participants in

both groups were still receiving treatment under the care of a health provider. 3 patients from MD group and 2 from OFP group discontinued care due to ineffectiveness of treatment. On the Global Rating of Change Scale, 81% from MD group and 70% from OFP group reported some level of improvement in involuntary movements (p-value 0.087). 57% from MD group and 80% from OFP group reported some level of improvement in their jaw pain (p-value 0.338) (Tables 5 & 6).

TMD Symptoms after getting treatment; within last 30 days of responding to survey:

All the participants in both groups were requested to fill out the six-item TMD Screener for the symptoms in the last 30 days irrespective of the onset of involuntary movements or pain. 9 participants in MD group and 4 participants in OFP group screened positive for TMD (p-value 0.880). There was no difference in average pain rating for jaw or temple pain for participants in both groups (p-value 0.592). The number of days participants noticed jaw or temple pain in the last 30 days, averaged 13 days in MD group compared to 2 days in OFP group (Welch test p-value 0.002, Mann-Whiney test p-value 0.185) (Tables 8 & 9). Even though the global score of JFLS was higher in both

groups, the difference was not statistically significant.

DISCUSSION

To our knowledge, this is the first study to compare OMD patients from MD and OFP clinics. We have summarized demographic features, clinical features particularly TMD related characteristics, general health status, and treatment outcomes from OMD patients' perspective from two different clinical settings in a single institution.

We tried to overcome the limitation of patient recall bias by asking several TMD related questionnaires (TMD pain screener, JFLS, characteristic pain intensity) in the last 30 days of completion of survey. Despite small sample size, both groups had comparable baseline and clinical characteristics. Demographic features like age and female prevalence noted in our study were similar to other published studies (1, 7). It was interesting to note all the participants that responded to our survey were Caucasians. It is unclear from literature if OMD is more prevalent in Caucasians (24) or if it is just a non-response bias.

Most of the studies measuring QoL in dystonia patients are based on cervical dystonia with very few studies on OMD patients (25) (11, 13, 26, 27). These

OMD studies have variably used Glasgow Benefit Inventory (GBI), oromandibular dystonia questionnaire-25 (OMDQ-25) or communication-related quality of life (CR-QoL) to measure quality of life after botulinum toxin treatment. We used generic health related Quality of Life measure [EuroQol (EQ)-5D-5L] as it also takes non-motor components (pain, anxiety, depression) of OMD into consideration. Further, it represents OMD participants' health at the time of completion of survey that makes it less susceptible to recall bias. Based on EQ-5D-5L summary score; OMD patients from both clinics did not reveal any significant difference. The result specific to OMD could be useful to compare disease burden with other diseases (e.g. diabetes, hypertension, cancer) across the globe (22).

Our study showed that 48% of OMD patients from MD clinic and 60% from OFP clinic reported jaw pain along with involuntary movements. In these patients with jaw pain, about 90% had positive TMD pain screener. Our findings parallel with Costa et al (8) where painful TMD was reported in 85% of OMD patients. It suggests that presence of TMD related characteristics is simply not a referral bias and the positive TMD pain screener could be due to overloading of the masticatory system with dystonic muscle contractions. Further, we tried to assess the temporal relationship of involuntary movements and jaw pain. Of the 16 participants

in both groups who reported pain, 50% reported onset of pain and involuntary movements around the same time while, 37% noted that involuntary movements preceded the pain. It is unclear at this time if jaw pain is a cause or effect of involuntary movements in OMD patients.

Botulinum toxin remained as the mainstay of care in OMD patients from both clinics as supported by evidence (11, 28). Clinical care in both clinics differed in terms of second line treatment provided to OMD patients i.e. oral medications (e.g. baclofen, deutetrabenazine, clonazepam) in MD clinic while oral appliance in OFP clinic. 24% (5/21) participants from MD group said medications provided the most relief which is similar to reported in literature (29). Several case reports have been published in reducing dystonic movements with oral appliances (30-32). 1 participant from each group reported oral appliance is the most effective treatment for involuntary movements. It remains questionable if medications or oral appliance is the choice of treatment in patients when Botox is contraindicated.

OMD patients in MD clinic noticed more improvement in their involuntary movements than jaw pain. Even after treatment, TMD pain screener remained positive in about 40% patients in both clinics in the last 30 days

questionnaire responses. Although the average jaw or temple pain was about the same in OMD patients from both clinics on graded chronic pain scale, the number of days with pain was more in MD group compared to OFP group. This suggests that treatment of TMD symptoms in addition to involuntary movements could lead to better patient outcomes.

The JFLS is a predictor of persistent TMD pain (33) and was found to be significantly higher in TMD cases than pain-free controls (34). JFLS global score was higher in OMD patients from both groups, which was similar to findings of Bakke et al (35). All these findings suggest that instead of considering TMD as a differential diagnosis of OMD (36, 37), we need to think of TMD as a co-existing feature along with involuntary movements in OMD.

Our study also highlights the feasibility of online platforms to conduct surveys. The survey response rate was 59%, higher than reported in literature (38). This could be attributed to additional reminders, monetary incentive upon completion of survey and easy access to this survey on handheld electronic devices after scanning QR code or typing short URL code on browser (39).

CONCLUSION

Our study demonstrates OMD patients presenting in two clinical settings do not seem to be different from each other in their clinical characteristics, quality of life and treatment outcomes by patient report. OMD patients from both clinics screened positive for TMD and reported higher scores on JFLS, suggesting TMD symptoms are a coexisting feature in patients with OMD.

.

Figure 1: Flow chart

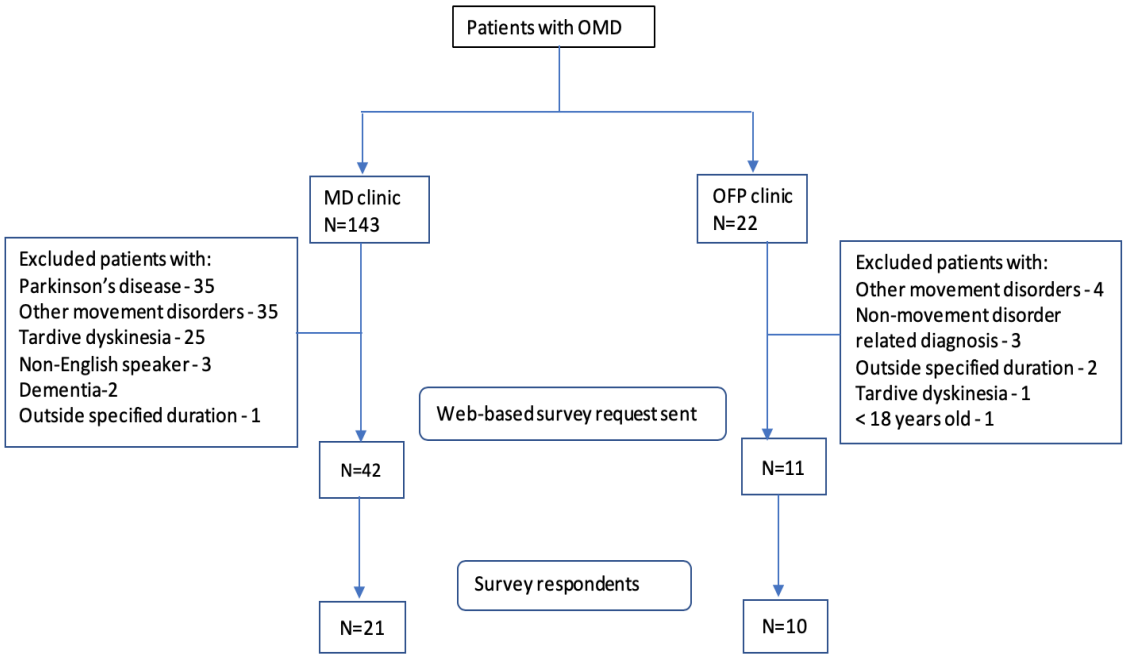

Table 1. Demographic questionnaires

Characteristics	N	MD clinic	OFP clinic	p-value
Q40. Age in years (mean, range) *	31	59.43 (31-82)	63.80 (55-84)	0.498
Q41. Sex – Female (n, %) ^	31	17 (81%)	6 (60%)	0.213
Q42. Race (n, %) ^	31	Caucasian (21, 100%)	Caucasian (10, 100%)	1
Q43. Level of education ^	31			0.775
High school graduate		3 (14.3%)	3 (30%)	
Some college/Associate degree		8 (38.1%)	3 (30%)	
Bachelor's degree		7 (33.3%)	3 (30%)	
Graduate Degree		3 (14.3%)	1 (10%)	
Less than high school		0 (0%)	0 (0%)	

* Mann-Whitney U test; ^ Chi-Square test

Table 2. General Health Status (EQ-5D-5L and general health question)

Survey Questions	N	MD Group	OFP group	P-value ^
Q34.Mobility				
No problems	31	13	9	0.223
Slight problems		4	1	
Moderate problems		4	0	
Severe problems		0	0	
Unable to walk about		0	0	
Q35.Self-Care				
No problems	31	20	10	0.483
Slight problems		0	0	
Moderate problems		1	0	
Severe problems		0	0	
Unable to wash/dress		0	0	
Q36.Usual activities (e.g. work, study, housework, family or leisure activities)				
No problems	31	14	9	0.347
Slight problems		5	1	
Moderate problems		2	0	
Severe problems		0	0	
Unable to do usual activities		0	0	
Q37.Pain/Discomfort				
No pain/discomfort	31	4	5	0.441
Slight pain/discomfort		11	4	

Moderate pain/discomfort		4	1	
Severe pain/discomfort		1	0	
Extreme pain/discomfort		1	0	
Q38.Anxiety/depression				
Not anxious/depressed	31	8	3	0.172
Slightly anxious/depressed		8	4	
Moderately anxious/depressed		5	1	
Severely anxious/depressed		0	2	
Extremely anxious/depressed		0	0	
Q39.In general, would you say your health is:				
Excellent	31	0	1	0.813
Very good		9	7	
Good		7	0	
Fair		5	1	
Poor		0	1	

^ Chi-Square test

Table 3. EQ-5D-5L Summary score

Group	N	Mean	SD	SE	p-value *
MD	21	0.789	0.214	0.047	0.374
OFP	10	0.855	0.131	0.041	

* Mann-Whitney U test

Table 4. Symptom description before getting treatment (including OMD features and TMD screener at the onset of involuntary movements)

Survey Questions	N	MD Group	OFP group	P-value
Q1. Do you or did you in the past have a diagnosis of oromandibular dystonia, which involves involuntary movements of your jaw, tongue and/or lips? ^				
Yes	31	19 (90%)	8 (80%)	0.416
No		2 (10%)	2 (20%)	
Q2. How long ago did you first start noticing involuntary movements? *				
Months	31	175.9	97.2	0.330
Q3. What do you think precipitated or triggered these movements? ^				
Orofacial trauma	31	1 (5%)	1 (10%)	0.300
Dental treatment		3 (14%)	3 (30%)	
Medications		3 (14%)	0	
Stress		2 (10%)	3 (30%)	
Other		3 (14%)	0	
Unknown		9 (43%)	3 (30%)	
Q4. What kind of involuntary movements do/did you have? (*check all that apply*)				
Jaw opening movement	31	9	3	N/A
Jaw closing movement		11	4	
Jaw deviation to side/s		8	3	
Forward jaw movement		2	2	
Tongue movements		6	4	
Lip movements		3	3	
Q5. How much did/do the involuntary movements bother you? ^				
A lot	31	9 (43%)	6 (60%)	0.313
A moderate amount		8 (38%)	4 (40%)	

A little		4 (19%)	0	
None at all		0	0	
Q6. Did/do you have any tricks to stop or reduce these movements? ^				
Yes	31	14 (67%)	5 (50%)	0.373
No		7 (33%)	5 (50%)	
Q7. What are/were the trick/s that you used to alleviate these movements?				
Free text; varied responses. Most common sensory trick – chewing or placing objects in mouth	19	5 out of 14	2 out of 5	N/A
Q8. Before receiving treatment for these movements, did/do the involuntary movements affect your ability to perform daily functions? ^				
Yes	31	15 (71%)	8 (80%)	0.610
No		6 (29%)	2 (20%)	
Q9. What functions did/does it affect? *(check all that apply)*				
Eating	31	12	6	N/A
Talking		11	5	
Swallowing		4	3	
Sleeping		6	3	
Other		5	1	
Q10. When the involuntary movements started, did you have any muscle tension or tiredness, or fatigue associated with your involuntary movements? ^				
Yes	31	15 (71%)	9 (90%)	0.248
No		6 (29%)	1 (10%)	
Q11. When the involuntary movements started, did you have any pain associated with your involuntary movements? ^				
Yes	31	10 (48%)	6 (60%)	0.519
No		11 (52%)	4 (40%)	

Q12. What was the association between your pain and involuntary movements, when you started noticing involuntary movements? ^				
Pain preceded your involuntary movements	16	1 (10%)	0	0.362
Involuntary movements preceded pain		3 (30%)	3 (50%)	
Both started around the same time		6 (60%)	2 (20%)	
There was no association between pain and involuntary movements		0	1 (10%)	
Q13. In the **30 days** when the pain started, how long did any pain last in your **jaw or temple** area on either side?				
From very brief to more than a week but it did stop	16	3	0	**See table 7**
Pain was always present		7	6	
Q14. In the **30 days** when the pain started, have you had **pain or stiffness** in your **jaw** on awakening?				
Yes	16	9	3	**See table 7**
No		1	3	

Q15. In the **30 days** when the pain started, did the following activities **CHANGE** any pain (that is, make it better or make it worse) in your **jaw or temple** area on either side?						
		MD group		**OFP group**		
		Yes	No	Yes	No	
Chewing hard or tough food	16	5	5	5	1	**See table 7**
Opening your mouth or moving your jaw forward or to the side		7	3	3	3	
Jaw habits such as holding teeth together, clenching, grinding or chewing gum		6	4	5	1	
Other jaw activities such as talking, kissing or yawning		4	6	3	3	
Q16. When the involuntary movements started, what bothered you the most? (select one) ^						
Involuntary movements	16	7 (70%)		3 (50%)		0.424

Jaw pain		3 (30%)	3 (50%)	
Q17.When the involuntary movements started, what made you seek health care for the first time? (select one) ^				
Involuntary movements	16	8 (80%)	3 (50%)	0.210
Jaw pain		2 (20%)	3 (50%)	

* Mann-Whitney U test; ^ Chi-Square test

Table 5. Clinical Care

Survey questions	N	MD group	OFP group	p-value
Q18.What types of health care providers have you sought care with for your involuntary movements? *(check all that apply)*				
Primary care physician	31	19	5	N/A
Emergency care or urgent care		3	6	
Neurologist, general		15	6	
Neurologist, movement disorder specialist		19	5	
Dentist, general		9	6	
Dentist, orofacial pain specialist		6	7	
Psychologist		6	7	
Physical therapist		9	5	
Chiropractor		6	0	
Other		6	2	
Q19.How many providers have you seen in total for this problem? *				
	31	7.7	4.9	0.231
Q20.What treatment/s did you receive for your involuntary movements? *(check all that apply)*				
Medication/s	31	12	4	N/A
Botulinum toxin injection/s		17	7	
Physical therapy		7	4	
Chiropractic care		4	1	
Self-care or homecare recommendations or behavior modification instructions		8	4	
Mouth guard/oral appliance		6	7	
Surgery		4	3	

Talk therapy or counseling		5	2	
Acupuncture		6	2	
Other		4	1	
Q21.Which treatment provided you the most relief with the involuntary movements? ^				
Medication/s	31	5	0	0.350
Botulinum toxin injection/s		10	4	
Physical therapy		0	1	
Chiropractic care		0	0	
Self-care or homecare recommendations or behavior modification instructions		0	0	
Mouth guard/oral appliance		1	1	
Surgery		2	2	
Talk therapy or counseling		0	0	
Acupuncture		0	0	
Other		3	2	
Q22.Are you still receiving treatment under the care of a health provider? ^				
Yes	31	16 (76%)	7 (70%)	0.713
No		5 (24%)	3 (30%)	
Q23.Why have you discontinued the treatment? ^				
Complete relief of symptoms	8	1	0	0.688
Treatment not effective		3	2	
Side effects from treatment		0	0	
Financially unaffordable		0	0	
Other		1	1	

* Mann-Whitney U test; ^ Chi-Square test

Table 6. Symptoms after getting treatment (Global rating of change scale, TMD screener at the time of response to questionnaire)

Survey questions	N	MD group	OFP group	p-value
Q24.With respect to your involuntary movements, how would you describe yourself now (after the treatment) compared to when your symptoms started? ^				
Worse (-5 to -1)	31	3	0	0.087
Unchanged (0)		1	3	
Recovered (1 to 5)		17	7	
Q25. With respect to your jaw pain, how would you describe yourself now (after the treatment) compared to when your symptoms started? ^				
Worse (-5 to -1)	31	3	0	0.338
Unchanged (0)		6	2	
Recovered (1 to 5)		12	8	
Q26. In the **last 30 days**, on average, how long did any pain last in your jaw or temple area on either side?				
From very brief to more than a week but it does stop	31	4	3	**See table 8**
No pain		11	7	
Pain is always present		6	0	
Q27. In the **last 30 days**, have you had **pain or stiffness** in your **jaw** on awakening?				
Yes	31	10	3	**See table 8**
No		11	7	
Q28. In the **last 30 days**, did the following activities **CHANGE** any pain (that is, make it better or make it worse) in your **jaw or temple** area on either side?				
		MD group	**OFP group**	
	31	Yes / No	Yes / No	**See table 8**
Chewing hard or tough food		7 / 14	4 / 6	

Opening your mouth or moving your jaw forward or to the side		7	14	2	8	
Jaw habits such as holding teeth together, clenching, grinding or chewing gum		7	14	3	7	
Other jaw activities such as talking, kissing or yawning		7	14	2	8	

^ Chi-Square test

Table 7. TMD screener 1 (participants who experienced jaw pain at the onset of involuntary movements)

TMD screener	MD group	OFP group	p-value ^
Positive (3 or >3)	9 (90%)	5 (83%)	0.696
Negative (<3)	1(1%)	1 (17%)	
Missing data	11	4	

^ Chi-Square test

Table 8. TMD screener 2 (all participants with involuntary movements at the time of response to questionnaire)

TMD screener	MD group (21 participants)	OFP group (10 participants)	p-value ^
Positive (3 or >3)	9 (43%)	4 (40%)	0.880
Negative (<3)	12 (57%)	6 (60%)	
Missing data	0	0	

^ Chi-Square test

Table 9A. TMD related questions

Survey questions	N	MD group	OFP group	p-value
Q29. In the **last 30 days**, ON AVERAGE how would you rate your jaw or temple pain? * That is, your usual pain at times you were in pain.				
	31	25	20	0.845
Q30. In the **last 30 days**, how many days have you had jaw or temple pain? *				
	31	12.5 days	1.5 days	0.185
Q31. Is there a time in the day that your jaw or temple pain typically starts or its worse?				
Upon awakening	31	9	2	N/A
In morning		8	1	
In afternoon		4	1	
In evening		6	4	
No pattern		5	3	

* Mann-Whitney U test

Table 9B. Global score of Jaw Functional Limitation Scale

Group	N	Mean	SD	SE	p-value *
MD	19	1.839	1.610	0.369	0.811
OFP	8	1.594	1.599	0.565	

* Mann-Whitney U test

Table 9C. Oral Behavioral Checklist

Group	N	OBC score 0 to 10	OBC score 11 to 20	OBC score 21 to 30	p-value ^
MD	21	14	7	0	0.445
OFP	10	8	2	0	

^ Chi-Square test

Bibliography

1. Nutt JG, Muenter MD, Aronson A, Kurland LT, Melton LJ, 3rd. Epidemiology of focal and generalized dystonia in Rochester, Minnesota. Mov Disord. 1988;3(3):188-94.
2. Joensen P. High prevalence of primary focal dystonia in the Faroe Islands. Acta Neurol Scand. 2016 Jan;133(1):55-60.
3. Asgeirsson H, Jakobsson F, Hjaltason H, Jonsdottir H, Sveinbjornsdottir S. Prevalence study of primary dystonia in Iceland. Mov Disord. 2006 Mar;21(3):293-8.
4. Balasubramaniam R, Ram S. Orofacial movement disorders. Oral Maxillofac Surg Clin North Am. 2008 May;20(2):273-85, vii.
5. Blanchet PJ, Rompre PH, Lavigne GJ, Lamarche C. Oral dyskinesia: a clinical overview. Int J Prosthodont. 2005 Jan-Feb;18(1):10-9.
6. Raoofi S, Khorshidi H, Najafi M. Etiology, Diagnosis and Management of Oromandibular Dystonia: an Update for Stomatologists. J Dent (Shiraz). 2017 Jun;18(2):73-81.
7. Slaim L, Cohen M, Klap P, Vidailhet M, Perrin A, Brasnu D, et al. Oromandibular Dystonia: Demographics and Clinical Data from 240 Patients. J Mov Disord. 2018 May;11(2):78-81.
8. Costa AL, Campos LS, Franca MC, Jr., D'Abreu A. Temporomandibular disorders in patients with craniocervical dystonia. Arq Neuropsiquiatr. 2011 Dec;69(6):896-9.
9. Sankhla C, Lai EC, Jankovic J. Peripherally induced oromandibular dystonia. J Neurol Neurosurg Psychiatry. 1998 Nov;65(5):722-8.
10. Page AD, Siegel L. Perspectives on the Psychosocial Management of Oromandibular Dystonia. Semin Speech Lang. 2017 Jul;38(3):173-83.
11. Bhattacharyya N, Tarsy D. Impact on quality of life of botulinum toxin treatments for spasmodic dysphonia and oromandibular dystonia. Arch Otolaryngol Head Neck Surg. 2001 Apr;127(4):389-92.
12. Charous SJ, Comella CL, Fan W. Jaw-opening dystonia: Quality of life after botulinum toxin injections. Ear Nose Throat J. 2011 Feb;90(2):E9.
13. Merz RI, Deakin J, Hawthorne MR. Oromandibular dystonia questionnaire (OMDQ-25): a valid and reliable instrument for measuring health-related quality of life. Clin Otolaryngol. 2010 Oct;35(5):390-6.
14. Jinnah HA, Factor SA. Diagnosis and treatment of dystonia. Neurol Clin. 2015 Feb;33(1):77-100.
15. Batla A, Stamelou M, Bhatia KP. Treatment of focal dystonia. Curr Treat Options Neurol. 2012 Jun;14(3):213-29.
16. Gonzalez YM, Schiffman E, Gordon SM, Seago B, Truelove EL, Slade G, et al. Development of a brief and effective temporomandibular disorder pain screening questionnaire: reliability and validity. J Am Dent Assoc. 2011 Oct;142(10):1183-91.
17. Von Korff M, Ormel J, Keefe FJ, Dworkin SF. Grading the severity of chronic pain. Pain. 1992 Aug;50(2):133-49.
18. Dworkin SF, Sherman J, Mancl L, Ohrbach R, LeResche L, Truelove E. Reliability, validity, and clinical utility of the research diagnostic criteria for Temporomandibular Disorders Axis II Scales: depression, non-specific physical symptoms, and graded chronic pain. J Orofac Pain. 2002;16(3):207-20.
19. Ohrbach R, Larsson P, List T. The jaw functional limitation scale: development, reliability, and validity of 8-item and 20-item versions. J Orofac Pain. 2008 Summer;22(3):219-30.
20. Ohrbach R, Markiewicz MR, McCall WD, Jr. Waking-state oral parafunctional behaviors: specificity and validity as assessed by electromyography. Eur J Oral Sci. 2008 Oct;116(5):438-44.

21. EuroQol Research Foundation. EQ-5D-5L User Guide. [Internet] 2019; Available from: https://euroqol.org/publications/user-guides.
22. Pickard AS, Law EH, Jiang R, Pullenayegum E, Shaw JW, Xie F, et al. United States Valuation of EQ-5D-5L Health States Using an International Protocol. Value Health. 2019 Aug;22(8):931-41.
23. Kamper SJ, Maher CG, Mackay G. Global rating of change scales: a review of strengths and weaknesses and considerations for design. J Man Manip Ther. 2009;17(3):163-70.
24. Gonzalez-Alegre P, Schneider RL, Hoffman H. Clinical, Etiological, and Therapeutic Features of Jaw-opening and Jaw-closing Oromandibular Dystonias: A Decade of Experience at a Single Treatment Center. Tremor Other Hyperkinet Mov (N Y). 2014;4:231.
25. Girach A, Vinagre Aragon A, Zis P. Quality of life in idiopathic dystonia: a systematic review. J Neurol. 2019 Dec;266(12):2897-906.
26. Weiss D, Hieber L, Sturm J, Bortlein A, Mayr I, Appy M, et al. Botulinumtoxin Improves both Generic and Disease-Specific Quality of Life in Cervical Dystonia. Front Neurol. 2017;8:561.
27. Page AD, Siegel L, Jog M. Self-Rated Communication-Related Quality of Life of Individuals With Oromandibular Dystonia Receiving Botulinum Toxin Injections. Am J Speech Lang Pathol. 2017 Jun 22;26(2S):674-81.
28. Sinclair CF, Gurey LE, Blitzer A. Oromandibular dystonia: long-term management with botulinum toxin. Laryngoscope. 2013 Dec;123(12):3078-83.
29. Teemul TA, Patel R, Kanatas A, Carter LM. Management of oromandibular dystonia with botulinum A toxin: a series of cases. Br J Oral Maxillofac Surg. 2016 Dec;54(10):1080-4.
30. Schneider R, Hoffman HT. Oromandibular dystonia: a clinical report. J Prosthet Dent. 2011 Dec;106(6):355-8.
31. Khan J, Anwer HM, Eliav E, Heir G. Oromandibular dystonia: differential diagnosis and management. J Am Dent Assoc. 2015 Sep;146(9):690-3.
32. Watt E, Sangani I, Crawford F, Gillgrass T. The role of a dentist in managing patients with dystonia. Dent Update. 2013 Dec;40(10):846-8.
33. Kapos FP, Look JO, Zhang L, Hodges JS, Schiffman EL. Predictors of Long-Term Temporomandibular Disorder Pain Intensity: An 8-Year Cohort Study. J Oral Facial Pain Headache. 2018 Spring;32(2):113-22.
34. Ohrbach R, Fillingim RB, Mulkey F, Gonzalez Y, Gordon S, Gremillion H, et al. Clinical findings and pain symptoms as potential risk factors for chronic TMD: descriptive data and empirically identified domains from the OPPERA case-control study. J Pain. 2011 Nov;12(11 Suppl):T27-45.
35. Bakke M, Baram S, Dalager T, Biernat HB, Moller E. Oromandibular dystonia, mental distress and oro-facial dysfunction-A follow-up 8-10 years after start of treatment with botulinum toxin. J Oral Rehabil. 2019 May;46(5):441-9.
36. Jankovic J. Etiology and differential diagnosis of blepharospasm and oromandibular dystonia. Adv Neurol. 1988;49:103-16.
37. Evatt ML, Freeman A, Factor S. Adult-onset dystonia. Handb Clin Neurol. 2011;100:481-511.
38. Guo Y, Kopec JA, Cibere J, Li LC, Goldsmith CH. Population Survey Features and Response Rates: A Randomized Experiment. Am J Public Health. 2016 Aug;106(8):1422-6.
39. Phillips AW, Reddy S, Durning SJ. Improving response rates and evaluating nonresponse bias in surveys: AMEE Guide No. 102. Med Teach. 2016;38(3):217-28.