Clinical evaluation of non-motor symptoms in jaw closing dystonia

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Abstract

Background The features of non-motor symptoms such as cognitive ability, psychiatric state, and quality of life (QOL) is unknown in patients with jaw closing dystonia. **Methods** Seven patients with jaw closing dystonia (2 males, 5 females; mean age 60.4 years) and 7 healthy age- and gender-matched subjects (mean age 54.1 years) participated in this study. The patients were examined to determine the location of pain and movement difficulties, with awareness of those measured using a numerical rating scale (0-10). Examinations of cognitive ability using Mini-Mental State Examination findings, psychological states using the Hospital Anxiety and Depression Scale and Hamilton Rating Scale for Depression, and symptoms using the Symptom Checklist-90-Revised were also conducted, as well as examinations of QOL using Oral Health Impact Profile and 36-Item Short Form Survey results. **Results** The awareness of pain and amount of movement difficulty varied among the jaw closing dystonia patients. Those patients also presented significantly lower scores for cognitive ability, and higher scores for depression and somatization as compared to the healthy subjects. In addition, the patients showed significantly modulated scores for QOL. **Conclusions** Non-motor symptoms affecting jaw closing dystonia patients may be prominent in regard to cognition, depression, and somatization, as well as QOL, while causal neuropathological relationships are also present.

Background

Oromandibular dystonia is a type of focal dystonia characterized by forceful contractions of the face, jaw, and/or tongue, which cause difficulties with opening and closing the mouth, and also often affects chewing and speech [1,2]. Oromandibular dystonia is known to be associated with dystonia of the neck muscles/spasmodic torticollis, eyelids, and larynx [3], with the combination of upper and lower dystonia sometimes termed cranial-cervical dystonia. When oromandibular dystonia occurs with blepharospasm, it may be referred to as Meige syndrome [4]. Despite the motor definition of primary dystonia, previous studies of dystonia patients have detected widespread abnormalities [5–8]. The purpose of this study was to clarify the clinical features of non-motor symptoms, such as cognitive ability, psychiatric state, and quality of life (QOL), in patients with jaw closing dystonia. Based on those results, neuropathological causality is also discussed. Furthermore, our findings may be clinically useful for distinguishing jaw closing dystonia from habitual teeth grinding and bruxism (primary bruxism) on the basis of elucidation of non-motor symptoms.

For the present investigation, we focused on dystonia with jaw closing dystonia considered to be secondary bruxism, as only apparent neurological disorder without a relationship to family history or secondary causes, such as drug exposure or disorders like Wilson's disease [9]. Our results are the first to demonstrate non-motor symptoms in patients with jaw closing dystonia.

Methods

Participants

Seven patients with jaw closing dystonia [2 males, 5 females; mean age ($\pm$ SD) 60.4 ± 16.1 years) who visited the Department of Neurological Dentistry of Nihon University Dentistry at Matsudo Hospital in order to undergo examinations and treatments were enrolled in this study. All patients were diagnosed with jaw closing dystonia by a neurological surgeon based on clinical examination findings, including computed tomography imaging in 1 and magnetic resonance imaging in 2 dystonia patients, and EMG results of jaw and neck muscle activities in all. Dental state was also examined, which revealed complete dentition in 5 and partially edentulous dentition in 2 patients who had partial dentures. Seven age- and gender-matched healthy subjects (staff members of Nihon University Dentistry at Matsudo) were also enrolled (mean 54.1 ± 11.8 years old). All of the healthy subjects had complete dentition and normal dental occlusion, and were free from pain and motor dysfunctions, with no complaints regarding the oromandibular, maxillofacial, head, neck, and shoulder regions, and without temporomandibular disorder (TMD) symptoms. Prior to the study, each of the patients and healthy subjects provided informed consent for participation according to the World Medical Association's Declaration of Helsinki. The study was approved by the Committee on Ethics of Nihon University School of Dentistry at Matsudo.

Experimental procedures

Each of the patients was examined for cognitive ability by a neurosurgeon at the medical clinic of our hospital. Following that medical examination, each was then further examined by 2 dentists in the neurological dentistry clinic to determine the location and intensity of pain and movement difficulties, as well as psychological and QOL state. Location as well as awareness of the intensity of pain and movement difficulties were examined using a numerical rating scale (NRS, 0–10 points), while cognitive ability was examined using the Mini-Mental State Examination (MMSE) [10]. Furthermore, psychological state was examined using the Hospital Anxiety and Depression Scale (HADS) [11], Hamilton Rating Scale for Depression (HRSD) [12], and Symptom Checklist–90-Revised (SCL–90R) [13]. QOL state was determined by use of the Oral Health Impact Profile (OHIP–49) [14] and Short Form Health Survey (SF–36) [15].

Awareness of pain and movement difficulty

All of the dystonia patients were examined in regard to the location of pain and movement difficulties, with the awareness of those determined using an NRS (0–10 points).

Cognitive ability

All of the patients and healthy subjects were examined in regard to cognitive ability using the MMSE [10], which is a widely used screening tool for diagnosis of both mild and major neurocognitive disorders. This examination analyzes 5 cognitive domains; orientation, memory, attention, language, and constructive

praxis. The MMSE questionnaire is comprised of 11 questions and obtained scores range from 0 to 30. In the present study, an MMSE cutoff score of 24 was used clinically to detect cognitive impairment [16].

Psychiatric states

Psychological problems and symptoms were examined using the HADS, HRSD, and SCL-90R for a broad range of psychological problems and symptoms related to psychopathology. The HADS [11, 17, 18] is a brief self-reporting two-dimensional questionnaire developed to screen for levels of anxiety and depression in patients, and one of the most widely used for those with neurological illnesses, and the HADS manual indicates that a score between 0 and 7 is “normal,” between 8 and 10 “mild,” between 11 and 14 “moderate,” and between 15 and 21 “severe.” [18]. The HRSD [12, 19] is a 21-question multiple choice questionnaire used for determining the severity of symptoms observed in relation to depression, such as low mood, agitation, anxiety, and weight loss. Each question has from 3–5 possible responses, which increase in severity. HRSD indicates no depression (0–7), mild depression (8–16), moderate depression (17–23), severe depression (≥ 24) [19]. The SCL-90-R [13, 20, 21] is one of the most widely used self-rating scales for considering psychopathological issues. Each item in this tool is measured in terms of how much the symptom is bothering or distressing the participant, and scored on a 5-point Likert scale ranging from 0 to 4. As for the time frame, this patient-self-reported questionnaire refers to the past week. For the present study, depression and somatization were evaluated using the SCL-90-R, and, normal is < 0.535 , moderate is $0.535–1.105$, and severe is >1.105 in depression, and normal is <0.5 , moderate is $0.5–1.0$, and severe is >1.0 in somatization [20].

QOL state

The OHIP-49 questionnaire [14, 22] was used to assess oral health-related QOL in the dystonia patients and healthy subjects. This questionnaire is comprised of 49 questions across 7 domains; functional limitations, physical pain, psychological discomfort, physical, psychological, and social disabilities, and handicap. The participants were instructed to rate the frequency of each OHIP-49 item on a 5-point Likert-like scale ranging from 0 to 4, with higher scores indicating more negative perceptions of the impact of oral condition on the sense of well-being. The patients and healthy subjects also reported perceived general health status using the SF-36 [15, 23], which measures the degree of feelings of disability in regard to daily activities. The survey is comprised of 36 questions divided into 8 scaled scores for vitality, physical functions, bodily pain, general health perception, physical, emotional, and social role functions, and mental health, which are subsequently transformed into a range from 0 to 100. Lower scores indicate higher levels of disability in daily activities.

Statistical analyses

T-test was used for comparing MMSE, HRSD, SCL–90R, HADS, OHIP–49, and SF–36 results between the dystonia patients and healthy subjects when normality was passed, while a Mann-Whitney rank sum test was used, when normality failed. All statistical analyses were performed using the SigmaStat software package, v. 3.11 (Systat Software, Inc, CA, USA) and results were considered to be significant when the value for comparison was less than 5%.

Results

Symptoms related to jaw closing dystonia

The main complaint noted by the dystonia patients was involuntary jaw clenching. Disease duration ranged from 0.5–2.5 years (average 2.0 ± 2.3 years). None of the patients had a history of medication for psychiatric issues or family history of dystonia. Involuntary muscle EMG activities were presented in the bilateral masseter, temporal anterior, anterior digastric, and sternocleidomastoid muscles in all 7 patients. Five of the patients had complete dentition, while 2 had partially edentulous dentition and wore partial dentures. Each of the healthy subjects had complete dentition.

Location of pain and movement difficulties, and NRS results

The patients had pain symptoms and movement difficulties that were not restricted to the jaw area, but were also noted in the head and shoulder regions beyond the facial area. In addition to jaw opening deficits, movement difficulties were also seen in regard to jaw closing, tongue movement, lip movement, and chewing and swallowing performances (Table 1). The level of awareness of pain and movement difficulties varied among the patients.

Cognitive ability

The patients had significantly ($p < 0.01$, Mann-Whitney rank sum test) lower scores for the MMSE as compared to the healthy subjects (Table 2).

Psychiatric state

HADS

The patients had significantly ($p < 0.05$, Mann-Whitney rank sum test) higher HADS scores for anxiety and depression than the healthy subjects (Table 2).

HRSD

In HRSD results, the patients showed significantly ($p < 0.01$, Mann-Whitney rank sum test) higher scores for depression than the healthy subjects (Table 2).

SCL–90R

In SCL–90R results as well, the patients presented significantly ($p < 0.05$, t-test) higher scores for depression and somatization as compared to the healthy subjects (Table 2).

QOL state

OHIP–49

The dystonia patients showed significantly ($p < 0.01$, t-test; $p < 0.01$, Mann-Whitney rank sum test) higher scores for the OHIP–49 in regard to functional limitations, physical pain, psychological discomfort, physical, psychological, and social disabilities, and handicap as compared to the healthy subjects (Table 3).

SF–36

In SF–36 results, the dystonia patients had significantly ($p < 0.01$, 0.05, t-test; $p < 0.01$, Mann-Whitney rank sum test) lower scores for functional function, physical roles, physical pain, general health, vitality, social function, emotional roles, and mental health than the healthy subjects (Table 4).

Discussion

This study was conducted to clarify non-motor symptoms, such as cognitive ability, psychiatric state, and QOL state, in patients with jaw closing dystonia. The patients examined in this study presented variable symptoms related to pain and movement difficulties beyond the orofacial region, while their awareness of those difficulties and motor deficits in the jaw, tongue, lip, face, and shoulder regions also widely varied.

Recently, [Gn](#) and [Nag](#) [24] reported clinical findings of a case of oromandibular dystonia in a 26-year-old female who complained of painful constrictive movements on the right side of the face and a feeling of constriction in the neck, which led to difficulty with breathing. Their patient experienced spontaneous, intermittent, unilateral paroxysmal, and severely painful involuntary spasmodic contractions on the right half of the face, which were repetitive throughout the day and could be relieved by conscious opening of the mouth, only to reappear again with the next occlusal contact. Considering the features reported in that study, it is conceivable that the dysfunctional awareness of pain and movement deficits may have great variety among individual jaw closing dystonia patients.

The jaw closing dystonia subjects in the present study had significantly decreased cognitive abilities as evaluated by MMSE findings. Romano et al. [25] reported that patients with cranial-cervical dystonia may have impaired cognitive domains related to working memory, processing speed, visual motor ability, and short term memory, while Yang et al. [8] also suggested that cognitive disabilities are predominant in cervical dystonia patients. In consideration of those previous reports of cognitive disabilities noted in dystonia patients, it is likely that those with jaw closing dystonia are also affected by cognitive decline,

similar to cranial and cervical dystonia patients [8, 25]. Additionally, the present patient cohort showed anxious and depressive psychiatric states. Clinical observations have noted psychiatric comorbidities, and frequent co-existence of depression and anxiety in dystonia patients [26, 27]. Thus, it is conceivable that jaw closing dystonia patients may also be affected by anxiety and depression as part of their psychiatric state.

Examinations of our patients with the SCL-90R also revealed aggravated somatization, which is considered to be psychological or emotional distress manifested in the form of physical symptoms related to a medically unexplained situation [28–30]. Mild sensory symptoms such as discomfort in the neck noticed months prior to the development of cervical dystonia, irritation or dry eyes prior to development of blepharospasm, and throat irritation prior to the development of spasmodic dysphonia have been reported [28, 31]. Associations of somatoform disorders with anxiety and depressive states have been reported in cervical dystonia patients [27, 32, 33], thus it is also assumed that jaw closing dystonia patients may develop a somatization state such as medically unexplained physical symptoms in relation to anxious and depressive states.

The present patients showed reduced QOL, based on their OHIP-49 and SF-36 scores. Zurowsky et al. [27] as well as others have confirmed that psychiatric illnesses have a major impact on QOL of patients with dystonia [34–36]. In our study, the dystonia patients consistently demonstrated lower QOL as compared to the healthy controls. Furthermore, OHIP-49 scores for the present patient cohort were consistent with those reported by Bakke et al. [37] for patients with Parkinson's disease. On the other hand, our patients had more severe SF-36 scores than reported for patients with cervical dystonia [38, 39], cranial dystonia [38], and writer's cramp [40, 41]. Interestingly, our jaw closing dystonia patients showed a remarkable resemblance to patients with Meige's syndrome in SF-36 results [42]. Taken together, we consider that jaw closing dystonia can have severe effects on physical QOL state, similar to Meige's syndrome.

Although the neuroanatomical basis of dystonia is unclear, the basal ganglia have been implicated in its pathophysiology based on observations of secondary dystonia in patients with basal ganglia lesions [28]. Structural lesions in the thalamus, parietal lobe, brainstem, and cerebellum can also cause secondary dystonia. In contrast, that study by Lipowski [28] showed that the absence of observable neurodegeneration in primary dystonia suggests an underlying neuronal dysfunction of connectivity, plasticity, and synaptic regulation involving the basal ganglia circuitry. Cortical-limbic-striatal dysfunction may be involved in depression and other neuropsychiatric disorders in dystonia patients, while attention-executive cognitive deficit may also develop in patients with young-onset generalized and adult-onset focal and segmental dystonia [7]. When considering neuropsychiatric factors [43, 44], sensory and cognitive function [45], and cognitive motor interactions [46] related to the basal ganglia, abnormal functioning basal ganglia may be involved in the pathogenesis of non-motor symptoms in dystonia patients, including jaw closing dystonia.

Bruxism, an abnormal repetitive movement disorder characterized by jaw clenching and tooth grinding, is classified as awake and sleep bruxism [47]. Awake bruxism is usually seen as a jaw clenching habit that appears in response to stress and anxiety states, and considered to be primary bruxism, while it does not have a relationship with other medical conditions. In contrast, neurological disorders such as jaw closing dystonia are termed secondary bruxism. In consideration of the clinical differences between awake bruxism and jaw closing dystonia, several similarities and dissimilarities were shown by the results of the present study. That is, awake bruxism [48, 49] and jaw closing dystonia present similar psychiatric states. In contrast, the jaw closing dystonia patients in our study also presented a dissimilarly marked decline of QOL state to a degree greater than that seen in TMD patients [50, 51]. Furthermore, when considering the strong correlation between exacerbation of TMD symptoms and diurnal bruxism previously reported [52], the oral and physical QOL outcomes of the present study may distinctly characterize jaw closing dystonia, and provide clues for clinical differentiation from awake bruxism patients. Our findings also indicate that cognitive disability might be associated with jaw closing dystonia [53, 54]. Taken together, it is speculated that non-motor symptoms, such as oral and physical QOL outcomes and cognitive disability, may be clinically applicable to differentiate patients with jaw closing dystonia from those with TMD with diurnal bruxism. In addition, though this study included 2 dystonia patients who were partially edentulous, it seems like partial edentulous status and denture wearing may not have a significant influence on QOL in jaw closing dystonia patients [55].

In summary, even though localization and awareness of pain, and movement difficulties are variable in patients with jaw closing dystonia, non-motor symptoms such as cognition, psychiatric state, and QOL may be prominently aggravated. Considering the pathogeneses of basal ganglia deficiencies, non-motor cognitive and psychiatric symptoms, as well as dystonic motor deficits may develop in association with jaw closing dystonia.

Conclusions

The present results clarify non-motor symptoms, such as cognitive ability, psychiatric state, and QOL, in patients with jaw closing dystonia. Modulated awareness of pain and movement difficulties were shown to be individually variable, while decreased and aggregated cognition, psychiatric states such as depression and somatization, and poor QOL in regard to oral condition and physical daily life were found. Considering the neural pathogenesis of dystonia, non-motor symptoms such as cognition, psychiatric state, and QOL, as well as motor deficit symptoms may also be prominent in affected patients with jaw closing dystonia.

Abbreviations

QOL: quality of life; TMD: temporomandibular disorder; NRS: numerical rating scale; MMSE: Mini-Mental State Examination; HADS: Hospital Anxiety and Depression Scale; HRSD: Hamilton Rating Scale for Depression; SCL-90R: Symptom Checklist-90-Revised; OHIP-49: Oral Health Impact Profile; SF-36: Short Form Health Survey

Declarations

Ethics approval and consent to participate

Ethics approval for the study was obtained from the Ethics Committee of Nihon University School of Dentistry at Matsudo. All participants gave their written informed consent.

Consent for publication

Not applicable.

Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

NN: Design and conceptualization of the study, statistical analysis and interpretation of data, drafting of manuscript; accountable for all aspects of the manuscript. TI: Acquisition of data, statistical analysis of data. KK: Acquisition of data, statistical analysis of data. TH: Interpretation of data, revision of manuscript for intellectual content. All authors read and approved the final manuscript.

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Tables

Table 1. Location of pain and movement difficulty, and numerical rating scale findings of pain and difficulty in 7 dystonia patients

atient	Location of pain	Awareness of pain (NRS) Comprehensive awareness	Location of movement difficulty	Awareness of movement difficulty (NRS)					
				Jaw closing	Jaw opening	Tongue movement	Lip movement	Chewing	Swallowing
a	Jaw	4	Jaw, lip	0	8	4	2	0	0
b	Jaw	5	Jaw	0	10	10	0	0	0
c	Jaw, mouth tongue, tooth, gingiva, shoulder	10	Jaw, tongue, face	10	10	6	10	10	10
d	Jaw, gingiva	10	Jaw, mouth, tongue	10	10	10	10	10	6
e	Jaw, mouth, tongue	6	Jaw, tongue, lip	4	4	3	8	5	2
f	Jaw, tongue, head, shoulder, eye	1	Jaw, tongue, lip, shoulder	2	3	3	2	3	2
g	Jaw, gingiva	8	Jaw, mouth	0	5	0	0	8	0

NRS Numerical rating scale

Table 2. Scores in the cognitive ability and psychiatric state in 7 healthy subjects and 7 dystonia patients

	Healthy subjects								Dystonia patients							
	A	B	C	D	E	F	G	Mean (SD)	a	b	c	d	e	f	g	Mean (SD)
MMSE	30	30	30	30	30	29	30	30 (0.0)	27	30	27	29	19	24	19	25 (4.2) ††
HADS																
Anxiety	6	0	4	6	5	5	5	4.4 (2.1)	4	9	13	15	13	16	0	10.0 (6.0) *
Depression	2	0	3	6	1	1	2	2.1 (2.0)	0	9	13	15	14	16	13	11.4 (5.5) †
HRSD																
	1	0	2	3	2	0	0	1.1 (1.2)	9	13	8	6	33	18	7	13.4 (9.6) ††
SCL-90R																
Depression	0.1	0	0	0.9	0.4	0	0	0.19 (0.32)	0.5	0.6	0.8	2	1.5	0.4	0.8	0.93 (0.58) **
Somatization	0	0	0.1	0.3	0.3	0	0	0.11 (0.16)	0	1.7	0.6	1.0	0.9	0.8	0.5	0.77 (0.51) **

**p<0.01, * p<0.05, t-test; †† p<0.01 Mann-Whitney rank sum test

Mini-Mental State Examination (MMSE), normal cognition (greater than or equal to 24), severe (≤ 9 points), moderate (10–18 points) or mild (19–23 points) cognitive impairment. Hospital Anxiety and Depression Scale (HADS), normal (0–7), borderline abnormal levels (8–10), abnormal levels (>11). Hamilton Rating Scale for Depression (HRSD), no depression (0–7), mild depression (8–16), moderate depression (17–23), severe depression (≥ 24). Symptom Checklist-90-Revised (SCL-90R) scale in depression, normal (< 0.535), moderate (0.535–1.105), severe (>1.105). SCL-90R scale in somatization, normal (<0.5), moderate (0.5–1.0), severe (>1.0).

Table 3. Scores in the Oral Health Impact Profile (OHIP) for 7 healthy subjects and 7 dystonia patients

	Healthy subjects								Dystonia patients							
	A	B	C	D	E	F	G	Mean (SD)	a	b	c	d	e	f	g	Mean (SD)
Functional limitation	3	0	0	3	3	0	0	1.3 (1.6)	14	31	29	36	18	28	15	24.4 (8.7) **
Physical pain	0	0	0	0	2	0	0	0.3 (0.8)	10	26	24	35	19	23	15	21.7 (8.1) ††
Psychological discomfort	0	0	2	0	0	0	0	0.3 (0.8)	8	20	19	19	9	15	4	13.4 (6.4) **
Physical disability	0	0	0	0	0	0	0	0 (0.0)	10	26	31	32	15	16	21	21.6 (8.4) **
Psychological disability	0	0	0	0	0	2	0	0.3 (0.8)	8	17	16	24	20	6	8	14.1 (6.9) **
Social disability	0	0	0	0	0	1	0	0.1 (0.4)	2	9	15	20	15	7	6	10.1 (6.3) **
Handicap	0	0	0	0	0	1	0	0.1 (0.4)	5	14	20	24	24	6	9	14.6 (8.2) **

** p<0.01, t-test; †† p<0.01, Mann-Whitney rank sum test

Table 4. Scores in the 36-item short-form health survey (SF-36) for 7 healthy subjects and 7 dystonia patients

Healthy subjects									Dystonia patients							
A	B	C	D	E	F	G	Mean (SD)	a	b	c	d	e	f	g	Mean (SD)	
Physical function																
90	95	90	95	100	80	100	92.9 (7.0)	54	54	54	18	43	51	-7.1	38.2 (23.8) ††	
Physical roles																
100	87	94	81	100	100	100	94.6 (7.6)	56	13	36	3	33	16	3	22.5 (19.7) **	
Physical pain																
100	100	74	100	100	84	100	94.0 (10.6)	62	17	50	17	40	31	40	36.8 (16.5) **	
General health																
77	82	42	67	87	62	82	71.3 (15.6)	47	30	50	38	38	25	47	39.0 (9.4) **	
Vitality																
81	63	31	63	88	75	81	68.8 (19.1)	56	31	47	18	40	27	40	37.0 (12.9) **	
Social functioning																
100	100	88	100	100	100	100	98.2 (4.7)	57	12	44	57	57	31	12	38.6 (20.5) ††	
Emotional roles																
100	83	75	83	100	75	100	88.0 (11.6)	56	14	44	6	35	19	6	25.7 (19.5) **	
Mental health																
80	80	60	80	90	75	75	77.1 (9.1)	55	25	38	22	49	27	38	36.5 (12.3) **	

** p<0.01, * p<0.05, t-test; ††: p<0.01, Mann-Whitney rank sum test