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ORIGINAL ARTICLE

Oromandibular dystonia screening questionnaire for differential diagnosis

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Abstract

Objectives Oromandibular dystonia, which is characterized by stereotypic, task-specific, or sustained contractions of masticatory and/or lingual muscles, is frequently misdiagnosed as temporomandibular disorders or psychogenic disease. Diagnostic delay in oromandibular dystonia is not acceptable; thus, a screening tool that can distinguish this condition from a temporomandibular disorder may be helpful for medical professionals unfamiliar with involuntary movements or temporomandibular disorders.

Materials and methods A questionnaire that included questions on the clinical features of oromandibular dystonia, such as stereotypy, task-specificity, sensory tricks, and morning benefit, and included questions to rule out temporomandibular disorders (total point range 0–40) was administered to 553 patients suspected to have involuntary movements.

Results Based on a careful examination and the differential diagnosis, the patients were divided into four groups: oromandibular dystonia ($n = 385$), oral dyskinesia ($n = 84$), psychogenic (functional) movement disorder ($n = 50$), and temporomandibular disorders ($n = 34$). The questionnaire had a high level of internal consistency as measured by the Cronbach's α (0.91), and item-total correlation was significant ($p < 0.001$). The test-retest reliability on two separate occasions showed a significant correlation ($p < 0.001$). Mean total scores of the questionnaire significantly differed among oromandibular dystonia (32.0), temporomandibular disorders (10.4; one-way analysis of variance, $p < 0.001$), oral dyskinesia (21.0; $p < 0.001$), and psychogenic (functional) movement disorder (13.7; $p < 0.001$).

Conclusions Findings of this study suggest that the present questionnaire is a simple diagnostic tool that is useful for tentative differentiation of oromandibular dystonia from temporomandibular disorders.

Clinical relevance This screening tool can be used to distinguish oromandibular dystonia from temporomandibular disorders.

Keywords Oromandibular dystonia · Questionnaire · Screening · Reliability · Temporomandibular disorders

Introduction

Dystonia is a movement disorder that is characterized by sustained or intermittent muscle contractions that cause abnormal, often repetitive, movements, postures, or both [1]. Dystonia is classified based on two main factors: clinical characteristics and etiology [1]. Oromandibular dystonia is a focal dystonia that is manifested on the masticatory, lower facial, and/or lingual muscles [2–5]. It clinically presents as jaw-closing dystonia, jaw-opening dystonia, jaw-deviation dystonia, jaw-protrusion dystonia, lingual dystonia, lip dystonia, or a combination of these abnormal movements [2–5] and interferes with chewing, swallowing, and speaking, resulting in social embarrassment and cosmetic disfigurement. The precise etiology of the condition remains unclear [6]. Oromandibular dystonia has been treated with various antispasmodic or anticholinergic agents, botulinum toxin injection [4, 7–9], muscle afferent block therapy [2, 3], splint therapy [10], and/or coronoidotomy [11, 12].

Unfortunately, oromandibular dystonia is often misdiagnosed as temporomandibular disorders, psychogenic disorder, bruxism, or disease of unknown etiology [2, 3, 5]. The temporomandibular joints are complex structures containing muscles, tendons, and bones. The masticatory muscles are primarily responsible for the movement of this joint. Temporomandibular disorders are characterized by craniofacial pain involving the joint, masticatory muscles, or muscle

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innervations of the head and neck. Diagnostic delay in focal dystonia has decreased over time; however, it is still not acceptable [13]. The average lag time between symptom onset and a final diagnosis of dystonia is between 3.8 [14] and 6.4 years [15]. Cervical dystonia, with a diagnostic delay of between 3.7 [16] and 6.8 years [17], is estimated to be more common than oromandibular dystonia, which suggests an even greater lag time.

Oromandibular dystonia must be a blind spot between medical science and dentistry. Although with a low frequency, neurologists, particularly movement disorder specialists, can diagnose temporomandibular disorder as oromandibular dystonia, i.e., bruxism as jaw-closing dystonia, and anterior disc displacement without reduction as jaw-deviation dystonia. Knowledge and experience of neurology and of dental or oral and maxillofacial surgery are needed for the differential diagnosis of oromandibular dystonia from temporomandibular disorders, which are practically impossible to obtain simultaneously. Thus, a simple diagnostic tool is needed to enable primary care physicians, neurologists, dentists, and oral surgeons to differentiate oromandibular dystonia from temporomandibular disorder and to initiate appropriate treatment rapidly. Taking into consideration the abovementioned facts, a screening questionnaire that would make it possible to tentatively diagnose oromandibular dystonia would be practically useful for medical professionals who are not familiar with involuntary movements or temporomandibular disorders. Furthermore, such a questionnaire must be clinically suitable for earlier diagnosis or possible reference to the appropriate experts.

The aim of this study was to confirm the reliability of a ten-item screening questionnaire in the differentiation of oromandibular dystonia from temporomandibular disorders and other movement disorders.

Materials and methods

Oromandibular dystonia screening questionnaire

This is a retrospective study which was conducted in a single institution and was based on the findings of the author. This questionnaire was developed and confirmed to be reliable on the basis of a standard method for health measurement scales [18] and also methods for rating scales for craniocervical dystonia described previously [19–22].

Item generation

Structured interviews were carried out by the author for each patient. Clinical features, all symptoms, medical history, and other relevant aspects of life that were adversely affected by symptoms were recorded precisely in the medical records. A preliminary questionnaire with 14 questions was prepared based on the author's 30 years of experience in treating both involuntary movements and temporomandibular disorders at both oral and maxillofacial surgery and neurology departments, and it was also based on information from the medical records. The preliminary questionnaire included questions about the patient's symptoms, the clinical features of oromandibular dystonia, such as stereotypy, task-specificity, sensory tricks, and morning benefit, and questions to rule out temporomandibular disorders.

Scale generation

The preliminary questionnaire was administered to 150 consecutive patients. Each question was scaled on a 5-point Likert scale representing an increasing severity of impairment: 0 = not at all, 1 = a little, 2 = moderately, 3 = quite a bit, and 4 = extremely.

A combination of exploratory factor analysis (principal components method with varimax rotation) and cluster analysis was applied for scale generation. In the subsequent item reduction phase, four of the original items were omitted because of low internal consistency with the other items or redundancy within a subscale. This resulted in a scale with three domains. A self-administered ten-item questionnaire for oromandibular dystonia (total point range 0–40) is shown in Table 1.

Domain 1: Symptom changes during sleep or stress (items 7, 8, 9, 10)
Domain 2: Task-specificity and sensory tricks (items 1, 2, 5, 6)
Domain 3: Stereotypy (items 3, 4)

The original version of this questionnaire was written in Japanese. The Japanese version was back translated into an English version (Table 1) by two professional English and Japanese translators. Japanese patients completed the Japanese version, while international patients were administered the English version.

The internal consistency determined by pairwise correlations among all items in the questionnaire in all combinations of possible pairs of the subscales was quantified by Cronbach's α [18]. A minimum α of 0.8 was regarded as the criterion. Test-retest reliability was evaluated in 58 untreated patients by intraclass correlation coefficients. To confirm the reliability of the questionnaire, the results were compared on two separate occasions.

Patients

Out of the patients who visited our department from 2014 to 2017 and had complaints of involuntary contracture or movement of the masticatory, lingual, and/or lower facial muscles, 569 patients (374 females and 195 males, mean age ± standard

Table 1 Ten-item questionnaire for oromandibular dystonia. The self-administered questionnaire includes questions concerning the clinical features of oromandibular dystonia, such as stereotypy, task-specificity, sensory tricks, and morning benefit

Have you experienced the following problems? Please mark one answer per question.

	Not at all	A little	Moderately	Quite a bit	Extremely
1. Do you experience involuntary contractions or movements in the mouth, jaw, tongue, or lips?	□	□	□	□	□
2. Do you have muscle contractions or movements that you are unable to control?	□	□	□	□	□
3. Is the region of the muscle contractions or movements (jaw, cheek, temple, tongue, or lips) always the same?	□	□	□	□	□
4. Is the direction of the muscle contractions or movements (mouth closing, opening, or tongue protrusion) always the same?	□	□	□	□	□
5. Do your symptoms appear only during a specific task (speaking, eating, opening mouth, etc.)? Or, was it so in the early phase when you noticed the symptoms?	□	□	□	□	□
6. When something is in your mouth (chewing gum, candy, or a mouth piece), or you touch your mouth or chin with hands or fingers, do the symptoms become milder? Or, was it so in the early phase when you noticed the symptoms?	□	□	□	□	□
7. Are you able to sleep without symptoms?	□	□	□	□	□
8. Are your symptoms less severe or absent in the morning and gradually worsen during the day? Or, was it so in the early phase when you noticed the symptoms?	□	□	□	□	□
9. Is the severity of your symptoms affected by tension or relaxation? Or, was it so in the early phase when you noticed the symptoms?	□	□	□	□	□
10. Is it difficult for you to control the symptoms when you are nervous or under stress?	□	□	□	□	□

deviation [SD] 53.4 ± 17.4 years) completed the final version of the questionnaire. Sixteen patients with tic, myokymia, akathisia, epilepsy, tremor, chronic pain, inflammation, and trauma were excluded from the analysis because the main purpose of this questionnaire was to differentiate oromandibular dystonia from temporomandibular disorders. Patients who were suspected to have degenerative, inherited, or other neurological diseases were referred to neurologists at our clinic. Oromandibular dystonia was diagnosed based on the characteristic clinical features of focal dystonia, such as a stereotyped pattern of muscle contraction, task-specificity, sensory tricks, overflow phenomenon, morning benefit, co-contraction, and the patients' electromyographic findings [2, 3, 5, 9, 10]. If a patient had more than four out of seven characteristics or findings (stereotyped pattern of muscle contraction, task-specificity, sensory tricks, overflow phenomenon, morning benefit, co-contraction, and electromyographic findings), the patient was diagnosed to have oromandibular dystonia. The patients were divided into four groups: oromandibular dystonia, temporomandibular disorder, dyskinesia, and psychogenic (functional) movement disorder. Oral dyskinesia refers to repeated, uncontrollable movements, such as licking of the lips or chewing-like movements [5]. Patients with psychogenic (functional) movement disorders had none of the aforementioned clinical features of focal dystonia; its characteristic features are inconsistent with the pattern, distribution, and velocity of the involuntary movements [23, 24]. Temporomandibular disorders involve a number of clinical conditions of the temporomandibular joint, masticatory muscles, and related structures. Temporomandibular disorders were diagnosed according to the diagnostic criteria for temporomandibular disorders (DC/TMD) [25].

The patients were examined for the clinical features of oromandibular dystonia and the associated subtypes or other dystonia, and they were asked for a history of consultations in the departments and previous diagnoses.

This study was performed in accordance with the Declaration of Helsinki under the approval of the institutional review board and ethics committee of Kyoto Medical Center.

Statistical analysis

One-way analysis of variance was used to compare the differences in the total scores among four groups. Post hoc Scheffé tests were performed to evaluate the differences. Pearson's correlation was used to analyze test-retest reliability and item-total correlation. All statistical analyses were performed using the statistical software package SPSS for Windows version 14.0 (SPSS Japan Inc., Tokyo, Japan). The null hypothesis was rejected at the 5% level ($p < 0.05$).

Results

Classification of patients

The questionnaire was administered to 553 patients who were suspected to have involuntary movement disorders. The patients were carefully diagnosed. If two conditions, for instance oromandibular dystonia and dyskinesia, or oromandibular

Table 2 Demographic characteristics of the four groups

	Total	Oromandibular dystonia	Oral dyskinesia	Psychogenic movement disorder	Temporomandibular disorders
Number of patients	553	385	84	50	34
Age (years) [mean (SD)]	53.4 (17.4)	50.9 (15.8)	69.8 (14.9)	50.8 (15.2)	45.8 (18.7)
Sex (female, male) [*N* (%)]	364 (65.9%), 189 (34.2%)	246 (63.9%), 139 (36.1%)	62 (73.8%), 22 (37.5%)	31 (62.0%), 19 (38.0%)	25 (73.5%), 9 (26.5%)
Duration of symptom (years) [mean (SD)]	3.6 (5.3)	3.8 (5.3)	2.2 (2.6)	4.4 (8.7)	3.3 (3.2)

dystonia and temporomandibular disorders, coexisted in the patients, they were classified based on their main and original entity. The patients were divided into four groups: oromandibular dystonia ($n = 385$), temporomandibular disorders ($n = 34$), dyskinesia ($n = 84$), and psychogenic (functional) movement disorder ($n = 50$). Seven patients in oromandibular dystonia group had secondary temporomandibular joint symptoms because of long-lasting dystonic muscle contraction. Eleven patients in the oromandibular dystonia group had mild oral dyskinesia. A patient with chronic pain related to temporomandibular disorders was classified into the temporomandibular disorder group. Patients' demographic characteristics are shown in Table 2.

Properties of the questionnaire

The questionnaire had a high-level internal consistency as measured by Cronbach's α (0.91). Test-retest reliability showed a significant correlation ($p < 0.001$). Item-total correlation was significant ($p < 0.001$) (Table 3). There were no floor effects; however, ceiling effects were observed in the stereotypy subscale (questions 1 and 2) (Table 3).

Total scores of the questionnaire significantly differed among oromandibular dystonia (32.0 ± 5.1) (one-way analysis of variance, $F [3, 549] = 395.9$, $p < 0.001$), temporomandibular disorders (10.4 ± 4.2) (one-way analysis of variance, $p < 0.001$), psychogenic movement disorder (13.7 ± 6.0) ($p < 0.001$), and oral dyskinesia (21.0 ± 4.6) ($p < 0.001$) (Fig. 1).

Clinical features of patients with oromandibular dystonia

The clinical characteristic features in oromandibular dystonia, such as observed stereotypy, task-specificity, sensory tricks, and morning benefit, are summarized in Table 4.

The average lag time between symptom onset and diagnosis was 3.8 years. The patients with oromandibular dystonia had consulted a mean of 4.1 departments or hospitals (range 1–30). The consulted departments included the departments of dentistry (67.3%), neurology (65.2%), oral and maxillofacial surgery (58.7%), psychiatry (33.5%), neurosurgery (28.8%), otorhinolaryngology (18.4%), acupuncture (18.2%), internal medicine (7.5%), orthopedics (4.2%), ophthalmology (3.5%), pain clinics (2.8%), rehabilitation (2.1%), plastic surgery (2.1%), and others (3.9%).

The patients with oromandibular dystonia were diagnosed with or were suspected to have conditions of unknown etiology (20.8%), temporomandibular disorders (19.4%), bruxism (14%), dystonia (12.2%), psychogenic disorders (9.8%), dyskinesia (9.1%), occlusal problem (3.6%), chronic pain (1.6%), or other conditions (3.5%), or they were considered to be normal (6.1%).

Discussion

The present study is the first to demonstrate the reliability of a screening questionnaire for patients with oromandibular

Table 3 Results of the reliability properties for the total score and for each domain

	Total score	Symptom changes during sleep or stress	Task-specificity and sensory tricks	Stereotypy
Number of items	10	4 (items 7, 8, 9, 10)	4 (items 1, 2, 5, 6)	2 (items 3, 4)
Mean score [mean (SD)]	32.0 (5.1)	11.8 (2.7)	12.2 (3.2)	7.8 (0.64)
Range	8–40	10–16	8–16	4–8
Internal consistency (Cronbach's α)	0.91	0.82	0.81	0.93
Item-total correlation	N/A	0.78	0.87	0.42
Scoring minimum [%]	0	0	0	0
Scoring maximum [%]	2.3	5.6	8.2	72.3

N/A not available

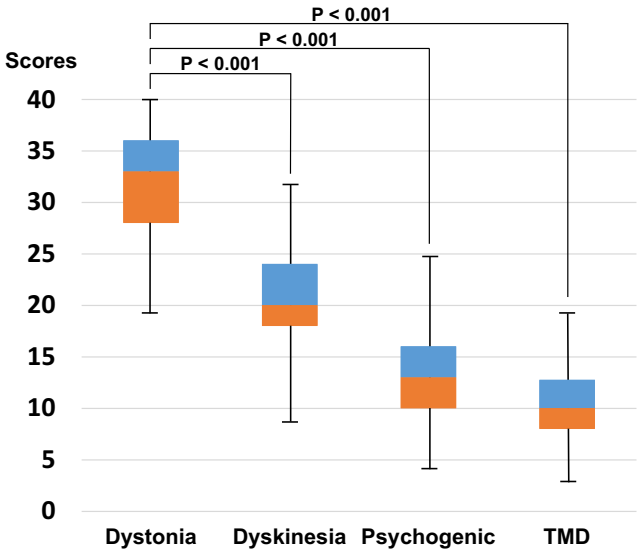

Fig. 1 Comparison of the results of total scores of the questionnaire among four groups. Total scores of the questionnaire significantly differed among oromandibular dystonia, temporomandibular disorders ($p < 0.001$), oral dyskinesia ($p < 0.05$), and psychogenic movement disorder ($p < 0.01$). Horizontal lines: median values, boxes 25th to 75th percentile; TMD temporomandibular disorders

dystonia. This study was based in one institution and the experience of one oromandibular dystonia specialist. All study patients were diagnosed, evaluated, treated, and followed by the same oromandibular dystonia specialist. Inconsistencies in the diagnosis were minimal. Significantly large differences in the total points of this questionnaire were observed between oromandibular dystonia and temporomandibular disorder groups. The mean total points of patients with oral dyskinesia and psychogenic movement disorder were intermediate to those of patients with oromandibular dystonia and patients with temporomandibular disorders. Naturally, further careful examination is needed for the precise diagnosis of each entity. Oral dyskinesia can coexist with oromandibular dystonia; some patients with oromandibular dystonia experienced secondary temporomandibular disorders due to long-lasting dystonic muscle contraction. Bruxism can coexist with oromandibular dystonia; such patients have muscle pain in the jaw-closing muscles on awakening. The questionnaire is useful for the tentative differentiation of oromandibular dystonia from temporomandibular disorders.

The present questionnaire was developed and confirmed to be reliable on the basis of a standard method for health measurement scales [18] and methods for rating scales for craniocervical dystonia described previously [19–22, 26]. The main purpose of this self-administered questionnaire was not to evaluate the severity of oromandibular dystonia, but to differentiate oromandibular dystonia from temporomandibular disorders. Hence, the content validity ratio, inter- and intraobserver reliability, discriminant validity, and convergent validity were not analyzed in this study. Strict prospective multicenter studies by multidisciplinary teams are required to confirm the reliability and validity of the questionnaire and to obtain a consensus. An obvious ceiling effect was observed in the "stereotypy" subscale (questions 3 and 4). However, stereotypy is the most typical clinical feature for focal dystonia. Therefore, the ceiling effect was a natural outcome, and thus, it is acceptable for the questionnaire. Nearly 90% of patients in this study had not been either diagnosed or suspected to have oromandibular dystonia. The patients visited 4.1 hospitals for 3.8 years to obtain the diagnosis of oromandibular dystonia. It is critical to diagnose patients

Table 4 Clinical characteristic features of patients with oromandibular dystonia

Number of patients	385
Associated movement disorders [N (%)]	
Cervical dystonia	46 (11.9%)
Blepharospasm	36 (9.4%)
Writer's cramp	12 (3.1%)
Spasmodic dysphonia	3 (0.8%)
Generalized dystonia	3 (0.8%)
Subtype of oromandibular dystonia [N (%)]	
Jaw-closing dystonia	229 (59.5%)
Lingual dystonia	98 (25.5%)
Jaw-opening dystonia	49 (12.7%)
Jaw-deviation dystonia	21 (5.5%)
Lip dystonia	14 (3.6%)
Jaw-protrusion dystonia	12 (3.1%)
Embouchure dystonia	3 (0.8%)
Stereotypy [N (%)]	369 (95.8%)
Task-specificity [N (%)]	269 (69.9%)
Speaking	184 (47.8%)
Chewing	63 (16.4%)
Mouth opening	14 (3.6%)
Swallowing	5 (1.3%)
Playing	2 (0.5%)
Singing	1 (0.3%)
Sensory tricks [N (%)]	198 (51.4%)
Chewing gum	81 (21.0%)
Touching with hand	35 (9.1%)
Candy	23 (6.0%)
Touching with finger	12 (3.1%)
Splint	12 (3.1%)
Handkerchief	10 (2.6%)
Mask	9 (2.3%)
Tooth pick	4 (1.0%)
Tissue paper	3 (0.8%)
Others	9 (2.3%)
Morning benefit [N (%)]	182 (47.3%)

promptly at the primary care level and refer them to appropriate experts. The questionnaire can be useful for early diagnosis. This simple screening questionnaire with ten questions might be useful as a self-check for patients who may consider visiting a clinic because of these symptoms. The questionnaire takes only minutes and can be filled out by a patient while in the waiting room on a first visit. The differentiation of this condition from temporomandibular disorders is important, particularly for the primary care physician, neurologist, and dental practitioner in the departments of dentistry or oral and maxillofacial surgery. A multidisciplinary team approach, involving clinicians who can diagnose temporomandibular disorders (oral and maxillofacial surgeon, temporomandibular joint specialist, or dentist) and physicians who can diagnose dystonia (neurologist, neurosurgeon, or psychiatrist), is preferable for diagnosis and treatment of oromandibular dystonia.

Each item of this questionnaire was intended to highlight the differences between oromandibular dystonia and other conditions, such as temporomandibular disorders, oral dyskinesia, and psychogenic movement disorders. Item nos. 1 and 2 (Table 1) are intended to check whether the respondents have involuntary contractions or movements.

Item nos. 3 and 4 (Table 1) are to confirm the stereotypy of the involuntary movements. Patients with oromandibular dystonia exhibit stereotypical jaw muscle contractions according to the subtype of oromandibular dystonia they have.

Item no. 5 (Table 1) is to check for task-specificity. Dystonia often starts with a specific task, especially in its early phase. The symptoms of oromandibular dystonia often appear task-specifically during speaking or chewing. The symptoms can later extend into other tasks and other body parts, and they might eventually be present at rest. The mean disease duration of oromandibular dystonia patients who exhibited task-specificity (3.0 years) was significantly shorter than that of patients without it (5.4 years) [10]. Therefore, the following note was added in the questionnaire: "Or, was it so in the early phase when you noticed the symptoms?" Additionally, the patients who did not already have this feature were required to check the item.

Sensory tricks can be assessed with question item no. 6 (Table 1). The sensory tricks are physical movements or positions that can temporarily interrupt dystonia, and patients may be aware of particular sensory tricks that provide some relief from their symptoms. The symptoms of oromandibular dystonia can sometimes be temporarily ameliorated via the use of these sensory tricks. Fifty-seven percent of patients with oromandibular dystonia had sensory tricks [23]. Sensory tricks are almost exclusive to dystonia and aid in its diagnosis. However, if a clinician has no knowledge of the phenomenon, the patient can easily be misdiagnosed as having a psychiatric disorder. Such patients may not even have recognized the sensory tricks despite actually using them [10]. The phenomenon can diminish after a long duration of the symptoms. Additionally, patients who did not have this feature were required to still check the item.

Item no. 7 (Table 1) distinguishes dystonia from bruxism as dystonic contractures are absent during sleep, whereas nocturnal bruxism is very common in temporomandibular disorders. This question is important for the differentiation of oromandibular dystonia from temporomandibular disorders.

The symptoms of dystonia tend to be milder in the morning (item no. 8), and this is described as morning benefit. These symptoms are in contrast to those of temporomandibular disorders caused by bruxism-related masticatory muscle tension, which tend to be worse upon awakening. This finding is also important for in the differential diagnosis.

Dystonic contractures are generally aggravated by stress (item nos. 9 and 10). On the other hand, the symptoms of temporomandibular disorder are not influenced directly by stress or tension. Patients cannot always respond to the questionnaire correctly. For instance, if taking a medication relieves the symptoms, a patient may check item 6 (sensory tricks). However, the medication is not a sensory trick. Furthermore, patients often do not notice the sensory tricks. Therefore, the questionnaire should be checked for its accuracy after completing the administration.

The prevalence of oromandibular dystonia is estimated to be 68.9 per million [27]. The number of patients with oromandibular dystonia was several dozens at most for a few decades, even in the studies by movement disorder centers or specialized neurologic departments. To the best of the author's knowledge, the present study evaluated the largest population of oromandibular dystonia patients. After the author launched a website focusing on involuntary movements in the stomatognathic system [28], more than 1000 patients from all over Japan and the world visited our clinic [29]. Most of them were misdiagnosed or were not adequately treated; therefore, they abandoned further consultation or treatment. The vast majority of patients with jaw-closing dystonia were diagnosed as having temporomandibular disorders or bruxism. The typical characteristic features, such as sensory tricks or task-specificity, can be misinterpreted to indicate a psychogenic disorder. The present questionnaire can be a practical screening tool even for medical professionals who are not familiar with involuntary movements or temporomandibular disorders.

Conclusion

This simple ten-item screening questionnaire is useful for tentative differentiation of oromandibular dystonia from temporomandibular disorders.

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Compliance with ethical standards

Conflict of interest The author declares no conflict of interest.

Ethical approval This study was performed in accordance with the Declaration of Helsinki under the approval of the institutional review board and ethics committee of Kyoto Medical Center.

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