Psychogenic Facial Movement Disorders: Clinical Features and Associated Conditions

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ABSTRACT: The facial phenotype of psychogenic movement disorders has not been fully characterized. Seven tertiary-referral movement disorders centers using a standardized data collection on a computerized database performed a retrospective chart review of psychogenic movement disorders involving the face. Patients with organic forms of facial dystonia or any medical or neurological disorder known to affect facial muscles were excluded. Sixty-one patients fulfilled the inclusion criteria for psychogenic facial movement disorders (91.8% females; age: 37.0 ± 11.3 years). Phasic or tonic muscular spasms resembling dystonia were documented in all patients most commonly involving the lips (60.7%), followed by eyelids (50.8%), perinasal region (16.4%), and forehead (9.8%). The most common pattern consisted of tonic, sustained, lateral, and/or downward protrusion of one side of the lower lip with ipsilateral jaw deviation (84.3%). Ipsilateral blepharospasm and excessive platysma contraction occurred in isolation or combined with fixed lip dystonia (60.7%). Spasms were reported as painful in 24.6% of cases. Symptom onset was abrupt in most cases (80.3%), with at least 1 precipitating psychological stress or trauma identified in 57.4%. Associated body regions involved included upper limbs (29.5%), neck (16.4%), lower limbs (16.4%), and trunk (4.9%). There were fluctuations in severity and spontaneous exacerbations and remissions (60%). Prevalent comorbidities included depression (38.0%) and tension headache (26.4%). Fixed jaw and/or lip deviation is a characteristic pattern of psychogenic facial movement disorders, occurring in isolation or in combination with other psychogenic movement disorders or other psychogenic features.

Key Words: facial movement disorders; psychogenic movement disorders; psychogenic facial movement disorders; psychogenic dystonia; psychogenic blepharospasm; facial distortion

Many systemic and neurological conditions may involve the facial musculature. From tetanus to blepharospasm, the majority of them are characterized by muscular spasms.1 While some of them are easily recognizable, anecdotal reports have recently focused attention on atypical presentations.2–5 Although most of these cases have been reported as representing rare phenotypes of organic focal dystonia,3–5 these patients may fulfill diagnostic criteria for psychogenic movement disorders (PMDs).
PMDs involving the face have been largely described as blepharospasm, reported in 3% to 7% of all types of PMD, and in 22% of a consecutive series of 50 patients in a botulinum toxin injection clinic. However, the features and diagnostic clues of the wider range of psychogenic facial movement disorders (PFMDs), as well as the relationship with previously reported cases in the Movement Disorders journal and others, remain unknown. We sought to examine a large series of PMDs where the orofacial region was involved in order to determine the clinical features and associated disorders, and to highlight their inconsistency and incongruence with recognized organic movement disorders.

Patients and Methods

Seven tertiary movement disorders centers performed a retrospective chart review of PMD involving the face using computerized databases and, when available, videotape examinations of patients evaluated between January 1993 and January 2010. Only patients actively followed up and treated by experts in movement disorders were included. Diagnosis of PMD was made according to the criteria of Fahn and Williams and Gupta and Lang. Patients were excluded when their clinical features were in keeping with those of hemimasticatory spasm or geniospasm or when they fulfilled established diagnostic criteria for dystonia (blepharospasm, oromandibular dystonia, Meige syndrome, or other dystonias, either focal or in the context of segmental or generalized forms), hemifacial spasm, or any neurological disease known to affect facial muscles (eg, myasthenia gravis, Parkinson’s disease, chorea, epilepsy). The following data were collected using a standardized spreadsheet: familial and demographic data,
clinical history, extent and type of facial and extra-facial involvement, neurophysiological investigations, type and outcome of the treatments performed by the movement disorders specialist (including psychotherapy and related techniques administered by other physicians upon request of the former) and, when available, long-term outcome data. Associated comorbidities and psychogenic features (including somatizations, false weakness or sensory complaints, suggestibility, or deliberate slowness of movements), secondary gain (ongoing or pending litigation, disability benefits, release from personal/legal/social/employment responsibilities, and/or increased personal attention), and disproportionate functional disability were also documented. Psychiatric diagnoses followed the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV TR) criteria. The diagnosis of a PFMD was not made on the basis of the presence or nature of associated psychiatric disturbances but on the clinical assessment of experienced movement disorders neurologists whose observations form the basis of this report.

Results
Demographics
The seven centers retrieved a total of 87 patients with PFMDs, representing the 16.3% ± 15.8% of all the PMDs cases seen during the examined period (range: 5.0% [Rome] to 50.0% [Boston]). Twenty-six cases were excluded because of missing data (eg, no video or limited follow-up) or uncertain clinical features, thus resulting in a total of 61 patients with PFMDs (92% females; mean age at onset, 37 ± 11.3 years; disease duration 6.7 ± 6.9 years; Table 1). They met published diagnostic criteria for PMDs (DSM-IV TR criteria for conversion disorder, 40.7%; somatization disorder, 9.8%; malingering, 4.9%).

Clinical Features
All patients had phasic or tonic muscular contractions resembling dystonia. The most prevalent feature was lip pulling (83.6%), predominantly downward (Fig. 1). Sixty percent had paroxysmal or intermittent

FIG. 1. The clinical presentation of PFMDs. The most common phenotype, isolated lower lip dystonia (A), in a patient with spontaneous remissions and intermittent ipsilateral jaw deviation (see also Video 1); eyelid spasm may be ipsilateral (B) or contralateral to the lip pulling (C) (see also Video 2). Note that the contraction of frontalis muscle involves the eyebrow contralateral to the spasm of the orbicularis oculi (arrows indicates a false "other Babinski sign"); platysma involvement is always associated with ipsilateral lip involvement (D), which can rapidly fluctuate in severity and appearance (E, F, same case); Some patients demonstrated severe bilateral spasms of most facial musculature (G), which may remit after placebo (vibrating tuning fork application, H) and relapse with different phenomenology shortly thereafter (I).
symptoms; fixed posturing was noted in most of the remainder. No patients acknowledged voluntary control or premonitory urge. Unilateral involvement was documented in most (84.3%), with alternating sides only in 2 subjects. There was lip pain and associated excessive ipsilateral platysma contractions in two-thirds of these.

Other clinical features are listed in Table 2.

Paroxysmal (65%) or fixed (26%) eyelid involvement occurred mostly unilateral with alternating sides (65%). The right side was affected twice as often as the left (11 vs 5). Three cases reported gestes antagonistes (1 at the second visit, after having received information on the phenomenon during the first visit). The frontalis muscle was often activated contralateral to the abnor-

### TABLE 2. The clinical features of patients with PFMDs with lip involvement

<table>
<thead>
<tr>
<th>N</th>
<th>51</th>
</tr>
</thead>
<tbody>
<tr>
<td>Involvement of</td>
<td></td>
</tr>
<tr>
<td>Any other facial muscle</td>
<td>24 (47.1%)</td>
</tr>
<tr>
<td>Platysma</td>
<td>37 (72.5%)</td>
</tr>
<tr>
<td>Neck, trunk, or limbs</td>
<td>22 (43.1%)</td>
</tr>
<tr>
<td>Onset outside face</td>
<td>5 (9.8%)</td>
</tr>
<tr>
<td>Facial/head pain</td>
<td>19 (37.2%)</td>
</tr>
<tr>
<td>Paroxysmal or action-induced only at onset</td>
<td>9 (17.6%)</td>
</tr>
<tr>
<td>Action-induced</td>
<td>3 (5.9%)</td>
</tr>
<tr>
<td>Paroxysmal symptoms</td>
<td>29 (66.9%)</td>
</tr>
<tr>
<td>Dystonic fixed posture</td>
<td>14 (27.5%)</td>
</tr>
<tr>
<td>Dystonic movement</td>
<td>40 (78.4%)</td>
</tr>
<tr>
<td>Consistency of laterality</td>
<td>40 (78.4%)</td>
</tr>
<tr>
<td>Asymmetry</td>
<td>43 (84.3%)</td>
</tr>
<tr>
<td>Side</td>
<td></td>
</tr>
<tr>
<td>Right</td>
<td>16 (31.4%)</td>
</tr>
<tr>
<td>Left</td>
<td>20 (39.2%)</td>
</tr>
<tr>
<td>Both asynchronous</td>
<td>7 (13.7%)</td>
</tr>
<tr>
<td>Both synchronous</td>
<td>8 (15.7%)</td>
</tr>
<tr>
<td>Direction of lip pulling</td>
<td></td>
</tr>
<tr>
<td>Upward</td>
<td>13 (25.5%)</td>
</tr>
<tr>
<td>Downward</td>
<td>32 (62.7%)</td>
</tr>
<tr>
<td>Both directions</td>
<td>3 (5.9%)</td>
</tr>
<tr>
<td>Sideways</td>
<td>3 (5.9%)</td>
</tr>
<tr>
<td>Type of speech</td>
<td></td>
</tr>
<tr>
<td>Normal</td>
<td>26/46 (56.5%)</td>
</tr>
<tr>
<td>Slurred</td>
<td>11/46 (23.9%)</td>
</tr>
<tr>
<td>Burst of verbal gibberish</td>
<td>7/46 (15.2%)</td>
</tr>
<tr>
<td>Stuttering</td>
<td>2/46 (4.3%)</td>
</tr>
<tr>
<td>Geste antagoniste (eg, placing in mouth a cigarette or a pencil)</td>
<td>3/49 (6.1%)</td>
</tr>
<tr>
<td>Effect of speech</td>
<td></td>
</tr>
<tr>
<td>No effect</td>
<td>28 (54.9%)</td>
</tr>
<tr>
<td>Improvement</td>
<td>8 (15.7%)</td>
</tr>
<tr>
<td>Worsening</td>
<td>15 (29.4%)</td>
</tr>
<tr>
<td>Effect of eating</td>
<td></td>
</tr>
<tr>
<td>No effect</td>
<td>33/41 (80.5%)</td>
</tr>
<tr>
<td>Improvement</td>
<td>6/41 (14.6%)</td>
</tr>
<tr>
<td>Worsening</td>
<td>2/41 (4.9%)</td>
</tr>
<tr>
<td>Effect of mouth movements (eg, kissing, whistling)</td>
<td></td>
</tr>
<tr>
<td>No effect</td>
<td>7/20 (35.0%)</td>
</tr>
<tr>
<td>Improvement</td>
<td>8/20 (40.0%)</td>
</tr>
<tr>
<td>Worsening</td>
<td>5/20 (25.0%)</td>
</tr>
<tr>
<td>Resolution during sleep</td>
<td>17/21 (80.9%)</td>
</tr>
</tbody>
</table>

PFMD, psychogenic facial movement disorder.

Progression and Outcome

The course was stable (53%) or variable (33%), with diurnal fluctuations in one-fifth. Spontaneous remissions were reported in 13 subjects (21%), with recurrence in 2 after 2 weeks and 10 years. Patients with or without remission did not differ with respect to number of visits, follow-up duration, gender distribution, age, occurrence of primary or secondary gain, resolution of pending medico-legal issues and disclosure of the diagnosis of psychogenicity to the patient. At the latest follow-up visit, spasms were present mostly in the lips (84%) and eyelids (51%). Isolated lip involvement was the most frequent pattern (43%), followed by lips and eyelids (23%) and eyelids alone (13%). Platysma was involved in 61% of patients. Extra-facial contractions involved upper limb (30%), neck (16%), leg (16%), and trunk (5%). When present, limb involvement was ipsilateral to the facial involvement. Dystonia was the most frequent phenotype of extra-facial sites (58%), followed by tremor (14%) and other jerks (10%). All but 6 subjects received a variety of medical and nonpharmacological treatments (Supplementary Table 1) without any benefit (56%) or worsening (20%). Aside from complete remissions (following a treatment or not), 20% of treated patients improved after treatment (Botulinum neurotoxin [BoNT] at therapeutic doses was effective in 5 cases, antidepressants in 3, antiepileptics in 2, and psychotherapy in 1).

Associated Conditions

Depression and headache were common (Supplementary Table 2A). Several features supported a psychogenic etiology (Supplementary Table 2B). Notably, most patients displayed variable phenomenology upon suggestibility maneuvers. A nonphysiologic or placebo maneuver (most often a vibrating tuning fork) improved 16% and worsened 10% of the 19 subjects to whom it was applied.

Discussion

This is the first series of PMD patients involving the face—exclusively or in association with other extra-facial movement disorders. A common clinical picture emerged, with asymmetric, mostly dystonic involvement of the lower face, resembling oromandibular dystonia, affecting predominantly young women (9:1 female-to-male ratio), and with a variety of features suggesting a psychogenic cause (Table 3). 2,6,7,9,11-16 Indeed, the vast majority of these cases received a
The unilateral involvement of facial muscles is common to PFMDs and hemifacial spasm (HFS). However, unlike the synchronous myoclonic jerks or tonic contractions, most patients with PFMDs show asynchronous, generally tonic contractions in ipsilateral lower and upper face or synchronous bilateral contractions of the lower face. In addition, we specifically looked for the “other Babinski sign” described in HFS patients with a specificity of 100% and characterized by ipsilateral eyebrow rising during eye closure due to the simultaneous contraction of orbicularis oculi and the internal part of the frontalis. This sign was not found in any of the patients with asymmetric spasm of orbicularis oculi, who, rather, had the eyebrow rising contralateral to the closing eye. Psychogenic HFS has been previously reported by Tan and Jankovic and was recently reviewed in the same center, where it accounted for 7.4% of all the cases referred for HFS. Almost all patients were women (15/16), mean age at onset and disease duration of symptoms were 37.4 ± 19.5 and 1.7 ± 2.2 years, respectively. Described patients had an acute onset of symptoms, a nonprogressive course, fluctuations in symptom severity, inconsistent signs, findings incongruent with HFS or facial dystonia, spontaneous improvement, and normal diagnostic studies. Facial spasm was characterized by upward or lateral deviation of the corner of the mouth; with bilateral involvement observed in 7 patients. Similar to our cases, no patients reported facial spasms during sleep (present in up to 80% of organic HFS) or worsening of spasms during voluntary facial contractions (documented in up to 39% of HFS patients); and most patients had lower-face involvement at onset in contrast to the isolated lid involvement typically present at onset in organic HFS. Interestingly, the report of a vascular loop compressing the seventh cranial nerve in 1 of our patients emphasizes the potential for this finding to be present in asymptomatic patients and the need for clinicians to recognize the important differences between HFS and PFMDs in order to avoid unnecessary diagnostic tests or treatments including surgical decompression. Electrophysiological examination may help in distinguishing HFS from other abnormal facial movements by demonstrating ephaptic impulse transmission between different facial nerve branches. Indeed, a neurophysiological hallmark of HFS is the spread of the blink reflex (BR) responses elicited by supraorbital nerve stimulation to muscles other than the orbicularis oculi. Interestingly, BR responses were normal when tested in 10 of our patients with PFMDs.

An uncommon involvement of the orbicularis oculi was demonstrated by 2 patients who could not open their eyes, a condition resembling “eyelid opening apraxia.” The strength of patients’ eye closure varied depending on the force exerted against the eyelids by the examiner. In some instances, the pattern of

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**TABLE 3. Features distinguishing organic versus psychogenic oromandibular and facial dystonia**

<table>
<thead>
<tr>
<th>Feature</th>
<th>Organic</th>
<th>Psychogenic</th>
</tr>
</thead>
<tbody>
<tr>
<td>Onset and progression</td>
<td>Gradual, slow progression</td>
<td>Sudden-onset, static course</td>
</tr>
<tr>
<td>Sensory tricks</td>
<td>May be present</td>
<td>Rarely present</td>
</tr>
<tr>
<td>Most common distribution</td>
<td>Bilateral</td>
<td>Unilateral</td>
</tr>
<tr>
<td>Most common site</td>
<td>Orbicularis oculi and frontalis</td>
<td>Orbicularis and frontalis, contralateral</td>
</tr>
<tr>
<td>Dystonic pattern</td>
<td>Phasic</td>
<td>Tonic</td>
</tr>
<tr>
<td>Dystonic exacerbation</td>
<td>Action-induced</td>
<td>Paroxysmal, maximum at rest</td>
</tr>
<tr>
<td>Dystonic spread</td>
<td>Segmental to cervical region</td>
<td>Segmental or multifocal</td>
</tr>
<tr>
<td>Evolution</td>
<td>Slowly progressive, no spontaneous exacerbations or remissions</td>
<td>Fluctuations in severity, spontaneous exacerbation and remissions</td>
</tr>
<tr>
<td>Pain</td>
<td>Usually absent</td>
<td>Present (25%)</td>
</tr>
</tbody>
</table>

*If orbicularis present in isolation, it most often occurred contralateral to the affected lip/jaw.

“positive” diagnosis rather than a diagnosis based on the exclusion of organic diseases.

Involvement of the lower lip with downward deviation at the angle of the mouth combined with ipsilateral platysma co-contraction, previously termed “smirk,” was the most frequent pattern at presentation. In contrast to the more common involvement of upper facial muscles in organic cranial movement disorders, the involvement of the lower face appears to be characteristic of PFMDs. Unlike organic oromandibular dystonia, most subjects had asymmetric facial involvement and absence of gestes antagonists. More importantly, the majority of patients had no involvement of speech, which, is commonly seen in oromandibular dystonia and has been reported in other series of PMDs. Moreover, although focal task-specific dystonias affecting perioral muscles are well described (eg, embouchure dystonia), “unilateral dystonia of the jaw” is uncommon and was first reported in 1986 by Thompson et al. in a small series. In retrospect, at least 1 of these patients was subsequently diagnosed as having a psychogenic etiology (J. Obeso, personal communication). Indeed, apart from masticatory spasm, whose clinical features are easily recognizable (eg, the association with facial hemiatrophy), few references to unilateral jaw spasms are to be found in the literature and several of these have clinical features that might support their reclassification as a PFMD (Table 4).
abnormal movement was very complex and not falling into any of the better defined movement disorders affecting the face. Examples included left blepharospasm with intermittent bilateral lower eyelid contractions associated with bilateral clonic movements of the levator labii superioris alaeque nasi muscle, or blepharospasm associated with movements of downward pulling of both angles of mouth followed by upward deviation and mouth opening. An important clue was provided by the ipsilateral contraction of the

<table>
<thead>
<tr>
<th>Reference (cases)</th>
<th>Onset/accompanied symptoms/precipitating events</th>
<th>Neurological examination</th>
<th>Natural history/response to treatment</th>
<th>Authors’ comments</th>
</tr>
</thead>
<tbody>
<tr>
<td>1(1) 14/F</td>
<td>Pain and tingling in the left mandible; chewing and talking difficulties</td>
<td>Tonic, sustained deviation of jaw to the left</td>
<td>No spreading; improvement with benzhexol</td>
<td>Focal dystonia of the jaw confirmed by: (A) presence of dystonia in other body parts (cases 2, 3, and 4); (B) continuous EMG activity of lateral pterygoid muscle at rest (case 1); (C) abnormalities in the recovery cycle of the blink reflex (case 3); (D) paroxysmal attacks of dystonia similar to those described in MS (case 5; normal imaging studies); and (E) improvement with anticholinergics</td>
</tr>
<tr>
<td>1(2) 30/F</td>
<td>Numberness on left side of face; tongue biting during the facial spasms</td>
<td>Torticollis and chin deviation to the left together after two years of forced and painful opening of the mouth</td>
<td>Spreading; partial benefit with benzhexol</td>
<td></td>
</tr>
<tr>
<td>1(3) 35/F</td>
<td>Dental extraction; chewing difficulties and tongue biting during the facial spasms</td>
<td>Sustained deviation of jaw to the right associated with intermittent ipsilateral torticollis</td>
<td>Transitory improvement with benzhexol and with amitriptyline thereafter</td>
<td></td>
</tr>
<tr>
<td>1(4) 22/F</td>
<td>No precipitating events reported</td>
<td>Intermittent left blepharoclonus and sustained deviation of jaw and lip to the left associated with ipsilateral torticollis and arm dystonia in the outstretched position</td>
<td>No relief with anticholinergics, carbamazepine, clonazepam, tetrabenazine</td>
<td></td>
</tr>
<tr>
<td>1(5) 28/F</td>
<td>No precipitating events reported</td>
<td>Right eye closure followed by upward deviation of the right corner of mouth; left eye adduction and spasms of orbicularis oris, mentalis, and left frontalis muscles; flexion and inversion of right foot while walking</td>
<td></td>
<td></td>
</tr>
<tr>
<td>22(1) 52/F</td>
<td>Phantom canine teeth and chronic facial pain after resection of hypertrophic gums</td>
<td>Tonic, sustained retraction of right corner of the mouth</td>
<td>No spreading; Improvement with doxepin and oxycodone</td>
<td>Peripheral injury induced dystonia</td>
</tr>
<tr>
<td>3(1) 40/F</td>
<td>Abrupt; associated with head and neck pain</td>
<td>Tonic, sustained, lateral and outward protrusion of the right lower lip</td>
<td>No spreading; No response to treatment; spontaneous improvement over time</td>
<td>Though unusual for primary dystonia (worsening at rest and improvement with labial movements) the stereotyped lip movements could not be entirely explained by a psychogenic cause</td>
</tr>
<tr>
<td>3(2) 41/F</td>
<td>Abrupt; accompanied by headache and left sided weakness</td>
<td>Tonic, sustained, lateral outward protrusion of the left lower lip</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3(3) 25/F</td>
<td>Abrupt; associated with headache and right sided weakness</td>
<td></td>
<td>Spreading to ipsilateral eyelid; spontaneous improvement over time</td>
<td></td>
</tr>
<tr>
<td>3(4) 42/F</td>
<td>Abrupt; accompanied with headache and left sided weakness</td>
<td></td>
<td>Lost to follow-up</td>
<td></td>
</tr>
<tr>
<td>4(1) 27/F</td>
<td>Abrupt; numbness in right cheek and right half of tongue</td>
<td>Tonic, sustained, lateral and outward protrusion of the right lower lip and right jaw deviation; present during sleep</td>
<td>Improvement with BoNT</td>
<td>Habit spasms superimposed on an abnormal facioctigeminal motor function after a Bell’s palsy</td>
</tr>
<tr>
<td>4(2) 39/F</td>
<td>Headache and numbness in right side of face</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5(1) 27/F</td>
<td>Acute onset after facial and trigeminal lesion secondary to a spider bite</td>
<td>Tonic, sustained downward deviation of left lower lip</td>
<td>Long-term improvement with a maxillary splint</td>
<td>Alteration of trigeminal input with secondary unbalanced inhibitory-excitatory activities within basal ganglia circuits</td>
</tr>
</tbody>
</table>

F: female; EMG, electromyogram; MS, multiple sclerosis; BoNT, botulinum neurotoxin.
platysma, a muscle uncommonly affected by organic movement disorders, with the exception of HFS, tics, and some cases of cranial dystonia, where it is usually bilateral. Interestingly, none of our cases reported ocu-
lomotor involvement (ie, oculogyria or convergence spasm), which has been recently reported in 13 PMDs
patients, 1 of whom had facial involvement. Finally, among patients with paroxysmal PFMDs, none
acknowledged voluntary control, urge, or relief of
urge after the movements, distinguishing these from
tics. Moreover, these patients never displayed rapid
movements and spasms were more sustained than
those observed in dystonic tics.

Fixed dystonia of the oromandibular region has been
reported to result from peripheral injury and may de-
velop within hours to months after dental procedures. This atypical cranial dystonia exhibits persistent pain
and dysesthesia, reminiscent of the limb causalgia syn-
drome, is also suggestive of a psychogenic etiology. Fixed
dystonic posture, in combination with sensory, autonomic, and motor disturbances, is a well recog-
nized manifestation of complex regional pain syndrome
(CRPS), reported as a complication of local trauma, of-
ten without peripheral nerve lesions. Although most
cases concern a limb, CRPS with focal dystonia can
involve muscles supplied by the cranial nerves as
well. The diagnostic dilemma of dystonia following
minor peripheral injury remains a major source of con-
trovery in this field given the psychogenic features
exhibited by these patients. Future electrophysiologi-
cal studies might help to unravel this controversy as BR
recovery cycle and sensorimotor plasticity have been
found normal in psychogenic blepharospasm and psy-
chogenic limb dystonia in contrast to the organic coun-
terparts. Nevertheless, it should be acknowledged that
electrophysiological studies might also disclose abnor-
mal findings in patients with PMDs.

Albeit commonly reported in PMDs literature, the
large preponderance of female gender in our sample is
in keeping with another series of PFMDs patients, thus
suggesting that women are particularly prone to
develop facial involvement in a context of psychoge-
nicity. Finally, in our sample, depression is the most
common associated condition with a prevalence by far
higher than observed in the same age group of women
in the general population (38% vs 4%). By contrast,
although very common, the prevalence of tension-type
headache is not higher than reported in the same pop-
ulation without PFMDs.

Our study has several limitations. The major short-
coming is represented by its multicenter retrospective
design, which is open to several errors (eg, missing
data, ascertainment errors, nonuniform modes of
assessment, recording data, and follow-up). However,
since the facial phenotype of PMDs has not been fully
characterized, its study could only arise from retro-
spective review rather than a prospective design. The
reliance on 7 centers employing the same standardized
data collection was intended to increase the reliability
of ascertainment of this rare phenotype and increase
the generalizability of the reported observations.
Moreover, the diagnosis of PMDs has been challeng-
ing because of historic transitions through the pitfalls
of underrecognition, misdiagnosis, overdiagnosis, and
diagnosis by exclusion. Furthermore, in the United
States, the management of PMD patients is hampered
by poor physician reimbursement, future insurability
of patients, and ongoing litigation. Although most
PMDSs (and also organic disorders such as Parkinson’s
disease), have no definitive “test” or “biomarker,” the
diagnosis can be reliably made by judicious application
diagnostic clinical criteria. Thus, it has been repeated-
ly emphasized in recent literature that this is not a di-
agnosis of exclusion but that positive features are
critical in making a diagnosis of a definite psychogenic
movement disorder. While in the past, cases of or-
ganic dystonia have often been mischaracterized as psy-
chogenic, many of us are impressed that the reverse
situation probably occurs more often now, with psy-
chogenic dystonia misdiagnosed as organic. Our report
is not only the first case series of psychogenic cranial
dystonia (and other movements) but also the first to
recognize as probably psychogenic some cases of disor-
ders affecting the oromandibular region previously
reported as organic, as discussed above. Despite this
large series of a relatively rare disorder (61 patients col-
lected by 7 tertiary-referral centers), it is possible that
the phenotypic range may still be larger than acknowl-
edged and biases of ascertainment and referral pattern
may have affected the results. Finally, the exclusion of
patients with organic movement disorders, precluded
our ability to address the coexistence of organic and
PMDSs, commonly seen in many patients.

In summary, PFMD should be considered when a
patient exhibits any combination of the following fea-
tures: (1) fixed or paroxysmal unilateral facial contrac-
tions, specially with lower lip with or without
epsilateral jaw involvement, of maximal severity at
onset; (2) inconsistent features such as changes in side
and pattern during or between examination; (3) asso-
ciated somatizations or nonphysiologic sensory or
motor findings; (4) reduction or abolition of facial
spasm with distraction; (5) response to suggestion or
psychotherapy; (6) rapid onset and/or spontaneous
remissions; and (7) normal neurological examination.
Supportive features are young age, female gender, and
associated medical conditions such as depression,
headaches, facial pain, fibromyalgia, or irritable bowel
syndrome. A prompt diagnosis based on phenomenol-
genic will avoid the extensive diagnostic workup char-
acteristic of a diagnosis-of-exclusion approach, prevent
unnecessary costly investigations, and permit
the institution of appropriate physical, psychological, and medical therapy.9

Legends to the Video

Video 1. This patient had sudden-onset right-arm weakness and fixed tonic muscular contraction affecting the right lower lip. Lip contraction attenuated when opening the mouth and protruding her tongue, and disappeared when laughing or trying to whistle, which functioned as a distracting maneuver. A “la belle indifférence” approach is noted. The expression at rest with improvement or disappearance with activity (“paradoxical dystonia”) is opposite to the typical action-induced expression of focal dystonias.

Video 2. Segment 1. Patient seen 5 months after sudden onset of fixed jaw deviation. Segment 2. Evaluation one month later, after sudden onset of contralateral blepharospasm. Distractibility with tongue movements and variability to passive manipulation of jaw opening and closing are demonstrated. Segment 3. Evaluation 1 year later demonstrates resolution of the blepharospasm but persistence of the fixed jaw dystonia.

References


30. Schrag A, Bhatia KP, Quinn NP, Marsden CD. Atypical and typical cranial dystonia following dental procedures. Mov Disord 1999;14:492–496.


