ABSTRACT: Psychogenic movement disorders are heterogeneous and diagnostically challenging. Despite the growing literature on adult forms, clinical features in children have received relatively little attention. We retrospectively reviewed medical records and video of patients <18 years diagnosed with a psychogenic movement disorder at our institute between 2007 and 2010. We identified 14 patients (6 males and 8 females) with a mean onset age of 11.5 years. Levels of diagnostic confidence were documented (2 patients), clinically established (8 patients), and probable (4 patients). A single movement disorder was present in 10 patients (71%); 4 patients (29%) presented an association of two or more movement disorders. Eleven patients presented other medically unexplained symptoms associated with their movement disorders. Five patients, among 6 with chronic occurrence, performed a polymyographic study showing significant modifications of frequency, amplitude, and distribution of electromyographic activity, related to distracting maneuvers. The present series represents 5% of all movement disorders observed in the considered period and 32% of nonorganic neurological manifestations. The most frequent movement disorders were tremor (36%) and dystonia (29%). We describe two phenotypes not previously reported among psychogenic movement disorders: myoclonus and association of myoclonus with dystonia. We remark on the presence of psychogenic symptoms associated with movement disorders (79%) as being one of the most useful clinical clues as well as on the value of polymyographic study in chronic psychogenic movement disorders, which provide evidence of the inconsistency of movement disorders. 

Key Words: psychogenic movement disorders; childhood

Psychogenic movement disorders (PMDs) are heterogeneous disturbances of motor function that are not explained by organic conditions and occur in association with underlying psychiatric disease and are diagnostically challenging.

Fahn and Williams proposed clinical and anamnestic criteria for the classification of PMDs in “documented,” “clinically established,” “probable,” and “possible” cases. The clinical usefulness of this classification was also underlined by Kirsch and Mink in children. Electrophysiological testing, in particular, polymyographic studies, was revealed to be useful in providing neuropathologic criteria to support the diagnosis of psychogenic myoclonus and tremor.

In movement disorder clinics, the frequency of PMD ranges between 2% and 4% for adults and between 2% and 3.1% for children. Despite the growing literature describing clinical features and natural history of adult patients with PMD, clinical characteristics of PMD in children have received relatively little attention.

Patients and Methods

We retrospectively reviewed medical records and video of all children (age, <18 years) included in our database under the term “psychogenic movement disorders.”
disorder” between 2007 and 2010. The movement disorder was classified by at least two movement disorder experts. Videotape recording was performed according to a standardized protocol: lying at rest (15 minutes), sitting at rest (15 minutes), speech (i.e., weekdays and months of the year), postural maintenance (i.e., arms and hands outstretched), finger-to-nose movements, rapid alternating movements of hands, voluntary movements (i.e., pouring water and threading beads), standing (15 minutes), walking (i.e., normal, tip-toe walk, tandem walk, backward walk, and running), and writing and drawing (e.g., Archimedes spiral).

Polymyographic study was performed in 3 patients. Electromyographic (EMG) activity was recorded with surface electrodes from at least one pair of antagonist muscles, which were chosen on the basis of clinical features of the patient. The study was performed at rest, in postural maintenance, during goal-directed movements, and during specific tasks (e.g., writing and drawing). EMG bursts (i.e., tonic activity and jerks or tremor) correlating with involuntary movements were measured in terms of amplitude, frequency, rhythmic or arrhythmic occurrence, duration, and synchronism or asynchronism on antagonist muscles. Modifications of EMG activity (e.g., frequency, amplitude, and distribution) related to distracting maneuvers (e.g., counting backward or performing rapid alternating movements of opposite limbs) were considered.

The level of diagnostic confidence of PMD was assessed according to Fahn and Williams criteria: “documented” (i.e., the movement disorder is persistently relieved by psychotherapy, suggestion, or placebo or disappeared when the patient is unobserved); “clinically established” (i.e., the movement disorder is incongruent or inconsistent plus other psychogenic signs, multiple somatisations, or obvious psychiatric disturbances); and “probable” (the movement disorder is incongruent or inconsistent, or psychogenic signs or multiple somatisations are present).

Results

We collected 14 patients (6 males and 8 females) with mean age at onset of 11.5 years (range, 5–17). Clinical features of the patients are summarized in Table 1.

According to Fahn and Williams criteria, the level of diagnostic confidence was documented in 2 (14%), clinically established in 8 (57%), and probable in 4 patients (29%).

In almost all patients (12 patients), the movement disorder started abruptly (86%), and in 2 cases, onset was subacute: 6 were chronic (43%), and 8 were episodic (57%), lasting from seconds to 5 hours with a great variability, even in the same patient.

All patients with chronic PMD had a static course, whereas in patients with episodic PMD, the duration of attacks tended to increase. A single movement disorder was present in 10 patients (71%); tremor in 4, myoclonus in 3, and dystonia in 3 patients). In 4 patients (29%), an association of two or more different movement disorders was found (myoclonus and dystonia in 2, tremor, myoclonus, and gait disturbance in 1, and dystonia and gait disturbance in 1 patient).

Cognitive profile was normal in 11 patients (78%); 2 presented with a mild cognitive impairment and 1 with a borderline IQ. All patients fit within the “somatoform disorder” category according to Diagnostic and Statistical Manual of Mental Disorders, 4th edition criteria. A psychiatric comorbidity was present in 7 cases (50%) and included anxiety, depression, panic attacks, and eating disorders.

In 11 patients (79%), there were other medically unexplained symptoms (i.e., psychogenic) variously associated with the movement disorders, represented by blurred vision, asthenia, fatigue, limb paresthesia, pain, dizziness, numbness, motor slowness, pseudoseizures, and unexplained falls; in 6, there was also a positive history for previous unexplained medical symptoms (e.g., recurrent pain, headache, sleep disturbances, and urinary urgency). Eight patients (57%) became so severely affected that they could not attend school.

Time from symptom onset until diagnosis of a PMD varied between 10 days and 10 years (mean, 20.5 months).

Five of six with chronic PMD (83%) performed a polymyographic study that documented tremor activity (cases 2 and 7), isolated myoclonic jerks (case 4), myoclonic jerks associated with tonic activity (case 8), and myoclonic jerks associated with tremor (case 14). In 4 of the 5 that performed the neurophysiological study, EMG activity was characterized by significant modifications, in terms of frequency, amplitude, and distribution, related to distracting maneuvers.

Seven of fourteen patients were followed after diagnosis for a period ranging between 6 months and 4 years: 3 cases fully recovered (1 with psychotherapy [case 14] and 2 spontaneously [cases 3 and 5]), and 4 remained chronically disabled (cases 1, 2, 9, and 12). No data are available for the other 7 patients.

Discussion

Neurological symptoms, not fully explained by organic diagnosis, are reported to occur from 9% up to 25% in different series of the adult population. Limited data are available in children: In a retrospective survey of 2,280 children with neurological
<table>
<thead>
<tr>
<th>Case no.</th>
<th>Gender, Onset Age (Years)</th>
<th>Mode of Onset</th>
<th>Movement Disorder</th>
<th>Occurrence and Course</th>
<th>Clinical Features</th>
<th>Psychiatric Comorbidity</th>
<th>Psychogenic Symptoms Associated</th>
<th>Level of Diagnostic Confidence</th>
<th>Time Lag Until Diagnosis</th>
<th>Polymyography</th>
<th>Cognitive evaluation</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>F, 15</td>
<td>Abrupt</td>
<td>Tremor</td>
<td>Episodic static</td>
<td>Generalized tremor, unable to attend school</td>
<td>—</td>
<td>Chronic musculoskeletal pain, urinary urgency</td>
<td>Clinically established</td>
<td>17 months</td>
<td>—</td>
<td>Normal</td>
</tr>
<tr>
<td>2</td>
<td>M, 8</td>
<td>Subacute</td>
<td>Tremor</td>
<td>Chronic static</td>
<td>Distal tremor at left upper limb, unable to attend school</td>
<td>—</td>
<td>—</td>
<td>Probable</td>
<td>10 years</td>
<td>Yes</td>
<td>Normal</td>
</tr>
<tr>
<td>3</td>
<td>F, 9</td>
<td>Abrupt</td>
<td>Myoclonus, dystonia</td>
<td>Episodic static</td>
<td>Dystonic posture of trunk and left upper limb, irregular, arrhythmic jerks at trunk and upper limbs, unable to attend school</td>
<td>—</td>
<td>Headache, pain, pseudoseizures, enuresis</td>
<td>Clinically established</td>
<td>3 years</td>
<td>—</td>
<td>Mild cognitive impairment</td>
</tr>
<tr>
<td>4</td>
<td>F, 15</td>
<td>Abrupt</td>
<td>Myoclonus</td>
<td>Chronic static</td>
<td>Irregular, arrhythmic jerks at left upper limb</td>
<td>Depression</td>
<td>Headache</td>
<td>Clinically established</td>
<td>10 days</td>
<td>Yes</td>
<td>Normal</td>
</tr>
<tr>
<td>5</td>
<td>F, 5</td>
<td>Abrupt</td>
<td>Myoclonus</td>
<td>Episodic static</td>
<td>Abdominal irregular, arrhythmic jerks</td>
<td>—</td>
<td>—</td>
<td>Clinically established</td>
<td>16 months</td>
<td>—</td>
<td>Normal</td>
</tr>
<tr>
<td>6</td>
<td>M, 14</td>
<td>Abrupt</td>
<td>Dystonia</td>
<td>Chronic static</td>
<td>Oromandibular and neck dystonia, unable to attend school</td>
<td>Depression, anxiety</td>
<td>—</td>
<td>Probable</td>
<td>4 months</td>
<td>—</td>
<td>Mild cognitive impairment</td>
</tr>
<tr>
<td>7</td>
<td>M, 17</td>
<td>Abrupt</td>
<td>Tremor</td>
<td>Chronic static</td>
<td>Generalized postural and action tremor, unable to attend school</td>
<td>Panic attacks</td>
<td>—</td>
<td>Documented</td>
<td>3 months</td>
<td>Yes</td>
<td>Normal</td>
</tr>
<tr>
<td>8</td>
<td>F, 17</td>
<td>Abrupt</td>
<td>Myoclonus, dystonia</td>
<td>Chronic static</td>
<td>Dystonic posture of upper limbs; multifocal irregular, arrhythmic jerks, unable to attend school</td>
<td>Depression</td>
<td>Abdominal recurrent pain, sleep disturbances, headache</td>
<td>Clinically established</td>
<td>8 months</td>
<td>Yes</td>
<td>Borderline</td>
</tr>
<tr>
<td>9</td>
<td>M, 8</td>
<td>Abrupt</td>
<td>Myoclonus</td>
<td>Episodic static</td>
<td>Irregular, arrhythmic jerks involving upper limbs and head</td>
<td>—</td>
<td>—</td>
<td>Clinically established</td>
<td>9 months</td>
<td>—</td>
<td>Normal</td>
</tr>
<tr>
<td>10</td>
<td>F, 9</td>
<td>Abrupt</td>
<td>Dystonia, gait disturbance</td>
<td>Episodic static</td>
<td>Dystonic movements and posture of head and neck at lower limbs, sometimes bilateral, and buckling of left knee</td>
<td>—</td>
<td>Headache</td>
<td>Clinically established</td>
<td>1 month</td>
<td>—</td>
<td>Normal</td>
</tr>
<tr>
<td>11</td>
<td>M, 11</td>
<td>Abrupt</td>
<td>Dystonia</td>
<td>Episodic static</td>
<td>Dystonic movements and posture of head and neck</td>
<td>Panic attacks</td>
<td>—</td>
<td>Probable</td>
<td>3 years</td>
<td>—</td>
<td>Normal</td>
</tr>
<tr>
<td>12</td>
<td>M, 15</td>
<td>Abrupt</td>
<td>Tremor</td>
<td>Episodic static</td>
<td>Lower limb tremor, then generalizing to upper limbs and abdomen, unable to attend school</td>
<td>Depression</td>
<td>Pain</td>
<td>Clinically established</td>
<td>15 months</td>
<td>—</td>
<td>Normal</td>
</tr>
<tr>
<td>13</td>
<td>F, 10</td>
<td>Abrupt</td>
<td>Dystonia</td>
<td>Episodic static</td>
<td>Dystonic facial movements</td>
<td>—</td>
<td>—</td>
<td>Probable</td>
<td>4 months</td>
<td>—</td>
<td>Normal</td>
</tr>
<tr>
<td>14</td>
<td>F, 11</td>
<td>Subacute</td>
<td>Tremor, myoclonus, gait disturbance</td>
<td>Chronic static</td>
<td>Upper limb postural tremor; irregular arrhythmic jerks at lower limbs with buckling of the knees, unable to attend school</td>
<td>Depression, eating disorder</td>
<td>Dizziness, pain</td>
<td>Documented</td>
<td>1 year</td>
<td>Yes</td>
<td>Normal</td>
</tr>
</tbody>
</table>

F, female; M, male.
disorders, a psychogenic etiology was reported in 6.4%. The present series represents 5% of all movement disorders and 32% of nonorganic neurological manifestations observed in the considered period at Istituto Nazionale Neurologico C. Besta (Milan, Italy), with those percentages being higher than what were previously reported in childhood literature.1,11,12,16

Fahn and Williams2 proposed different levels of diagnostic confidence for PMDs. Other investigators3,6 then suggested clues that may be useful for the diagnosis of psychogenic origin: “historical” clues (e.g., abrupt onset, static course, psychiatric comorbidity, and presence of multiple somatisations); “clinical” clues (e.g., incongruence, inconsistency, false sensory findings, distractibility, variability, and episodic occurrence); and “treatment” clues (e.g., remission with psychotherapy or placebo). The level of diagnostic confidence of our patients was documented in 14%, clinically established in 57%, and probable in 29%. The useful clues, suggesting the psychogenic origin, did not differ from what was reported in literature: among treatment clues, we were able to document remission in 2 patients with psychotherapy and placebo, respectively. Nevertheless, as underlined by Lang and Voon,18 very few historical and clinical clues suggesting psychogenic origin are completely specific, and only one study has attempted to evaluate sensitivity and specificity of proposed diagnostic criteria.19 Surely, the diagnosis should rely heavily on the presence of positive support at clinical examination, such as inconsistency and incongruency with the organic movement disorder counterpart. These features highlight the need for considerable clinical experience, both with PMD and nonpsychogenic movement disorders. Moreover, the diagnostic role of psychosocial factors in PMD has been recently put in argument.20

Below are some comments on the peculiar features of our series.

We found an earlier onset age, with 38% of patients under the age of 10, in contrast to what has been reported in the literature (13%),12,17 with 1 case with onset at 5 years.

The majority of patients (10 patients) of our series had a generalized movement disorder, and only 4 cases had a segmental form involving the nondominant side. This is in contrast with childhood PMD literature,12 which reports a more frequent involvement of the dominant side.

Unlike other series,1,12 no clear precipitant factors or stressor events have been reported in our patients, except for 1 case with a minor teeth surgery.

Medically unexplained symptoms, associated with PMD, are reported to be present in 40% to 91% of cases.1,12 We found psychogenic symptoms associated with PMD in 11 patients (79%) of our series, with pain being the most frequent complaint (diffuse pain or headache in 8 patients), followed by episodic visual symptoms (e.g., blurred vision, visual disperception, or dysopia in 3 patients) and asthenia and fatigue in 3 patients.

Psychiatric comorbidity is reported to be present in 13% to 52% of patients with PMD.1,12 We found it in 7 patients of our series (50%), and it was mainly represented by depression (5 patients), concordant with what was previously reported by Ferrara et al.,1 followed by panic attacks (2 patients).

Family history has been reported to be related to phenomenology of psychogenic MD; Ferrara and Jankovic1 reported 11% of PMD modeling. In 43% of cases (6 patients), there was a positive family history for neuropsychiatric disorders (e.g., multiple sclerosis, schizophrenia, anxiety, migraine, and cerebral tumor), but none of our patients showed a clear PMD modeling.

Unnecessary surgeries, procedures, and medications for PMD or for associated symptoms eventually determined to have no identifiable organic basis have been described1; we found 8 patients (57%) that received medications for the presumed organic movement disorder (e.g., benzodiazepines, antiepileptics, and baclofen).

Time from symptoms to diagnosis varied largely (between 10 days and 10 years; mean, 20.5 months), concordant with what was previously reported in the literature.12 This is the expression of the complexity of the diagnosis, but considering that the prognosis is considerably better in patients with short duration of symptoms,16,18 the need for early diagnosis is crucial.

According to the literature, the most common PMDs in children are dystonia (47%), tremor (40%), and gait disorders (13%).16 In our series, the most frequent movement disorder was tremor (36%), followed by dystonia (29%). In 2 cases, we found gait disturbances associated with tremor or dystonia. We describe two phenotypes not previously reported among PMD: myoclonus involving the upper limbs, abdomen, and/or head in 3 patients (21%; cases 4, 5, and 9) and the association of myoclonus with dystonia in 2 patients (14%; cases 3 and 8). In 1 patient (case 8), the distribution of dystonia and myoclonus was consistent with the phenotype of classic DYT11 dystonia.

Psychogenic dystonia is relatively frequent; its diagnosis is usually reported as the most challenging because it is often “fixed,” eliminating helpful diagnostic clinical features such as variability, distractibility, increase with attention, and entrainment.18 Nevertheless, this is referred to adults, whereas psychogenic dystonia in children is not fixed and is usually diagnosed on the basis of inconsistency and variability.

Polymyography has been a supportive criterion for psychogenic origin in 4 of 5 patients, showing variability, in terms of frequency, amplitude, and distribution, related to distracting maneuvers as well as absence of cocontraction and overflow. However,
these features are not always easily detectable in children, requiring a proper setting that includes prolonged registration to verify distractibility and extensive electrode displacements to verify overflow. On the other hand, cocontraction may be present in voluntary dystonic movement, as observed in patient 14.

We have few data on the outcome of our patients because half of them have not been followed after diagnosis: 21.5% recovered fully and 28.5% remained chronically disabled.

Schwingenschuh et al.\textsuperscript{12} reported on the outcome of pediatric PMD in their series of 15 cases (47% recovered fully, 33% improved substantially, and 20% remained chronically and severely affected) and in 50 cases reviewed from the literature (74% recovered fully, 14% improved partially, and 8% remained chronically disabled, and in 4% outcome was not reported). Brief duration of symptoms and presence of tremor as movement disorder are reported as good prognosis factors\textsuperscript{12,18} whereas no differences are reported in onset age, comorbidity, stressors, or precipitants.

**Conclusion**

In conclusion, we would like to remark on the presence of psychogenic symptoms associated with movement disorders (79%) as being one of the most useful clinical clues of our series as well as on the value of polymyographic study in chronic PMD, which provide evidence of the inconsistency of movement disorders.

**Legends to the Video**

**Video 1. Segment 1.** Case no. 7: generalized tremor with great frequency variability and disappearing of the movement disorder during particular motor tasks, such as writing and self dressing. **Segment 2.** Case no. 3: abnormal dystonic posture of the trunk and left upper limb with irregular jerks involving the trunk and upper limbs; 4 months after diagnosis, complete disappearance of the movement disorder. **Segment 3.** Case no. 14: irregular jerks of lower limbs and buckling of the knees.

**References**