Psychogenic Movement Disorders in Children: A Report of 15 Cases and a Review of the Literature

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Abstract: Data on psychogenic movement disorders (PMD) in children are scarce, with most existing literature relating to adults only. We report 15 cases with the aim of highlighting the clinical characteristics, risk factors, comorbidity, treatment, outcome, and prognosis of PMD in children. Only 13% of cases had onset before age 10, with the mean age at onset being 12.3 years. Females were predominantly affected (F:M = 4:1). The most common types of movement disorders seen were dystonia (47%), tremor (40%), and gait disorders (13%). Multiple hyperkinetic phenomenologies were observed in many cases. Abrupt onset and precipitation by minor injuries, and stressful life events were commonly reported. Clinical clues on examination suggesting a psychogenic origin were similar to those identified in adults. A distinct feature of PMD in children was the predominant involvement of the dominant limb. The underlying psychiatric diagnosis was conversion disorder in the majority of cases. Time from symptom onset until diagnosis of a PMD varied broadly (between 2 weeks and 5 years). Treatment with cognitive and behavioral therapy and rehabilitation by a multidisciplinary team led to improvement in most cases. However, treatment was much more effective in children with a short time from symptom onset to diagnosis and treatment.

Psychogenic movement disorders (PMD) are diagnostically and therapeutically challenging to both neurologists and psychiatrists. In adults they can mimic the whole range of movements seen in movement disorders of organic origin. In a series of 152 adult patients with PMD the most common were dystonia (62%), tremor (16%), gait disorder (11%), and myoclonus (8%).1 Diagnostic criteria for PMD have been published2 and include abrupt onset, inconsistency, incongruency, distractibility, false weakness, false sensory signs, pain, exhaustion, excessive startle, bizarre movements, and concomitant somatizations among others. Factors supportive of the diagnosis are selective disability, other psychogenic features on examination, history of a precipitating event, disease exposure (usually a family member), possible secondary gain, and comorbid psychiatric disease.3–6

Despite the growing literature describing clinical features and natural history of adult patients with PMD, the clinical characteristics of PMD in children have received relatively little attention. We therefore undertook a retrospective study of 15 children diagnosed with PMD to delineate their clinical characteristics, risk factors, psychiatric comorbidity, treatment, and prognosis.

PATIENTS AND METHODS

We retrospectively reviewed the medical records of all children (age ≤18 years) who were diagnosed with
RESULTS

From our clinics we identified sufficient information on 15 children. According to published diagnostic criteria, a documented PMD was the diagnosis in six, whereas the remaining nine were categorized as clinically established.

The psychiatric diagnosis showed conversion disorder in 12 and somatization disorder in 3 children. Demographics and clinical characteristics are summarized in Table 1.

Mean age at onset was 12.3 years (range 7–16 years; SD: 2.6 years). Only two cases presented before the age of 10, with the youngest age at onset being 7 years; 80% were female (F:M = 4:1). Fourteen of 15 children were right-handed. Only two children had a positive family history of a movement disorder (Parkinson’s disease, tics). Seven children had preexisting organic or psychiatric comorbidity.

In all the children the movement disorder started abruptly. One girl with dystonia progressed over several years, but all others reached the maximum severity of symptoms within a few days to months. In eight cases a clear stressor was elicited prior to the onset of symptoms. The onset of the movement disorder occurred within 2 weeks of a minor injury in six children (40%); in one of the children, after tonsillectomy and in one after being badly bullied in school. In the remaining seven (47%), no specific precipitating event was found. Attention-seeking behavior (secondary gain) was suspected in 20% and a mechanism to avoid a potentially difficult social/psychological situation in 40%.

The most common movement disorder was dystonia (n = 7). Among children with dystonia, the initially affected body part was the neck in three, the right arm in two, the right leg in one, and the left leg in one. Dystonia was paroxysmal in two children and continuous with fluctuations in the remaining children. In five patients dystonia spread within a few weeks to involve other body parts. Dystonia was fixed in 71% and was accompanied by pain in 57%. The distal part of the limb was most commonly affected. No patient had a geste antagoniste.

The second most common movement disorder was tremor (n = 6). In all six children with tremor, the right (dominant) hand was affected. Tremor was episodic in three cases. The tremor in all patients was characterized by a combination of variability, distractibility, entrainability, absent finger tremor, and exacerbation with stress and attention.

In two children, a gait disorder was the predominant feature. They presented with an unusual bizarre gait that was variable, inconsistent, and incongruent on examination.

In six children (40%), PMD had a single phenomenology (dystonia in 2 and tremor in 4), while the remaining nine children exhibited multiple motor phenotypes. Other medically unexplained symptoms were present in 40%. Approximately half of the children became so severely affected that they could not attend school.

Time from symptom onset until diagnosis of a PMD was on average 9.4 (SD 15.1) months. Extensive investigations including imaging, electrophysiological tests, levodopa challenge, genetics, plasma copper and ceruloplasmin, plasma amino acids, and urinary organic acids were performed in all the children except those with tremor. Following diagnosis of PMD, most children were looked after by a multidisciplinary team and treated with a combination of psychotherapy and physiotherapy (in one child the parents refused the possibility of a psychogenic etiology and treatment). Only three children were treated with an antidepressant (amitriptyline).

We defined remission as sustained (>1 year) disappearance of the PMD. After an average follow-up of 3.1 years, six children were in remission, six had improved substantially and could return to a normal school life, and three remained chronically and severely disabled without significant improvement. Patients with complete remission had a significantly shorter disease duration until the correct diagnosis was made (mean 0.7 years; SD 0.2 years) compared with those without improvement or with only partial improvement (mean 15.2 years; SD 17.4 years; t-test, P = 0.03). In our cohort, patients with tremor were most likely to remit (5 of 6) compared with those with gait disorder (1 of 2) or dystonia (0 of 7) as the predominant PMD. None of the patients taking an antidepressant recovered fully. Patients with full remission compared with those without any remission did not differ significantly with respect to age at onset, comorbidity, stressor or precipitant, and treatment.
<table>
<thead>
<tr>
<th>G/dominant side</th>
<th>AO</th>
<th>MO</th>
<th>Known stressors</th>
<th>Precipitant</th>
<th>Comorbidity</th>
<th>Main MD</th>
<th>Clinical characteristics</th>
<th>Treatment before diagnosis of PMD</th>
<th>Time lag until outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>F/R</td>
<td>16</td>
<td>Abrupt</td>
<td>Parents separated; half sister with Down’s syndrome</td>
<td>Fall during ice skating 2 days before onset, minor arm injury</td>
<td>Anxiety disorder</td>
<td>Dystonia, fixed</td>
<td>Fixed posture of R arm; progressed; to involve the leg; belle indifference; Pain; shaking attacks; unable to attend school</td>
<td>Co-codamol, Diclofenac, Levodopa, Benzhexol, Temazepam</td>
<td>8 mo/amitriptyline, physiotherapy, rehabilitation, psychotherapy/chronically disabled</td>
</tr>
<tr>
<td>F/R</td>
<td>14</td>
<td>Abrupt</td>
<td>Learning difficulties</td>
<td>None</td>
<td>Cerebellar hypoplasia, learning difficulties</td>
<td>Dystonia fixed</td>
<td>Paroxysmal head tilt to the R; anarthria, walking difficulties; unable to attend school</td>
<td>Levodopa, Benzhexol, Clonazepam</td>
<td>5 mo/physiotherapy, psychotherapy/relapsing remitting course; could return back to school</td>
</tr>
<tr>
<td>F/L</td>
<td>12</td>
<td>Abrupt</td>
<td>None</td>
<td>Tonsillectomy 2 weeks before onset</td>
<td>None</td>
<td>Dystonia</td>
<td>Episodic and progressive dystonia of neck; L hand; fixed dystonia legs; needed wheelchair; clearly worsening under emotional stress</td>
<td>Various analgesics</td>
<td>9 mo/OT, psychotherapy, psychotherapy/amitriptyline/partial recovery</td>
</tr>
<tr>
<td>F/R</td>
<td>7</td>
<td>Abrupt</td>
<td>Parents divorced 6 mo before</td>
<td>None</td>
<td>None</td>
<td>Dystonia</td>
<td>Attacks of severe fixed dystonia of L leg; pain in sacral area and acute urinary retention; needed wheelchair; inconsistent, incongruent; belle indifference</td>
<td>Levodopa (initial response with recovery for 6 mo), benzhexol, sodium-valproate, botex Carbamazepine, ibuprofen</td>
<td>5 yr/amitriptyline, physiotherapy, psychotherapy/chronically disabled</td>
</tr>
<tr>
<td>F/R</td>
<td>9</td>
<td>Abrupt</td>
<td>None</td>
<td>Fall backwards, minor neck injury</td>
<td>None</td>
<td>Dystonia</td>
<td>None</td>
<td>Co-codamol, Diclofenac, Levodopa, Benzhexol, Temazepam</td>
<td>9 mo/parents refused possibility of a psychogenic diagnosis and treatment/chronically disabled</td>
</tr>
<tr>
<td>M/R</td>
<td>15</td>
<td>Abrupt</td>
<td>Grandfather admitted to hospital; stung by a jellyfish on R foot 1 week before onset</td>
<td>Often headaches; delayed puberty, receives testosterone injections</td>
<td>Dystonia</td>
<td>Paroxysmal and fluctuating dystonia in R foot, shoulder, face, also chest, triggered by stress and attention, inconsistent. High anxiety</td>
<td>Levodopa (response for 2 weeks)</td>
<td>2 weeks/benzodiazepines, baclofen/fully recovered</td>
<td></td>
</tr>
<tr>
<td>F/R</td>
<td>10</td>
<td>Abrupt</td>
<td>None</td>
<td>Was tripping over in school, minor injury to R arm before onset</td>
<td>None</td>
<td>Dystonia and freezing</td>
<td>Fixed and painful dystonia R arm, freezing episodes of gait, sometimes whole body freezing</td>
<td>Analgesics</td>
<td>19 mo/CTB, psychotherapy/partial improvement</td>
</tr>
<tr>
<td>F/R</td>
<td>14</td>
<td>Abrupt</td>
<td>Bullied at school</td>
<td>Head and facial, obsessive features</td>
<td>Tremor</td>
<td>R-hand tremor; inconsistent and incongruent; more marked when attention is brought to it</td>
<td>Levodopa (response for 2 weeks)</td>
<td>1 mo/psychological support/fully recovered</td>
<td></td>
</tr>
<tr>
<td>F/R</td>
<td>15</td>
<td>Abrupt</td>
<td>Disabled due to organic disease</td>
<td>Moderate learning difficulties cerebellar malformation, ataxia, cadaveric renal transplant</td>
<td>Tremor</td>
<td>R-hand tremor, disappeared with distraction, variable, entrainable</td>
<td>Analgesics</td>
<td>2 weeks/psychotherapy, CBT/fully recovered</td>
<td></td>
</tr>
<tr>
<td>Gender/dominant side</td>
<td>Age at onset (AO)</td>
<td>Mode of onset (MO)</td>
<td>Known stressors</td>
<td>Precipitant</td>
<td>Comorbidity</td>
<td>Main MD</td>
<td>Clinical characteristics</td>
<td>Treatment before diagnosis of PMD</td>
<td>Time lag until diagnosis/treatment/outcome</td>
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<tr>
<td>F/R</td>
<td>10</td>
<td>Abrupt</td>
<td>None</td>
<td>None</td>
<td>None</td>
<td>Tremor</td>
<td>Episodic tremor of R arm lasting 2 to 3 days, Irregular, distractable, entrainable</td>
<td>3 weeks/psychotherapy, CBT/fully recovered</td>
<td></td>
</tr>
<tr>
<td>F/R</td>
<td>12</td>
<td>Abrupt</td>
<td>None</td>
<td>After a judo bout</td>
<td>None</td>
<td>Tremor</td>
<td>Tremor R arm; variable, distractable, entrainable; weakness and numbness R arm and leg</td>
<td>1 mo/CTB/fully recovered</td>
<td></td>
</tr>
<tr>
<td>F/R</td>
<td>11</td>
<td>Abrupt</td>
<td>Teased and bullied at school</td>
<td>None</td>
<td>None</td>
<td>Tremor and jerks</td>
<td>Episodic tremor and slow and irregular jerks R arm, precipitated by stressful situations (go to school). Episodes disappeared in holidays</td>
<td>12 mo/psychotherapy, physiotherapy/partial improvement</td>
<td></td>
</tr>
<tr>
<td>M/R</td>
<td>12</td>
<td>Abrupt</td>
<td>None</td>
<td>None</td>
<td>Mild tic disorder</td>
<td>Tremor</td>
<td>Episodic tremor R arm (usually at school); variable, distractable, entrainable</td>
<td>3 mo/CTB/fully recovered</td>
<td></td>
</tr>
<tr>
<td>F/R</td>
<td>15</td>
<td>Abrupt</td>
<td>None</td>
<td>Prior transverse myelitis</td>
<td>Gait disorder</td>
<td>Bizarre gait; imbalance with no falls; belle indifference; variable lower limbs weakness; pseudoseizures</td>
<td>1 yr/ongoing psychiatric care; physiotherapy/partial improvement</td>
<td></td>
<td></td>
</tr>
<tr>
<td>F/R</td>
<td>12</td>
<td>Abrupt</td>
<td>None</td>
<td>Minor head injury</td>
<td>Gait disorder</td>
<td>Bizarre bouncing gait; Variable, inconsistent, incongruent, intermittent pseudoseizures</td>
<td>2 weeks/physiotherapy and CBT/fully recovered</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

G, gender; AO, age at onset; MO, mode of onset; R, right; L, left; M, male; F, female; MD, movement disorder; OT, occupational therapy; CBT, cognitive behavioral therapy; mo, month; yr, year.
DISCUSSION

Given the scarcity of data regarding phenomenology, outcome, and therapy, PMD in children and adolescents pose a difficulty from both a diagnostic and management point of view. Here we discuss the findings of our 15 cases of pediatric PMD in the context of 50 previously published cases retrieved from literature. We extracted information regarding gender, age, and mode of onset, phenomenology of the movement disorder, relevant background history, investigations, treatment, and outcome from the following references and the number of cases from each reference are as shown: n = 5; n = 7; n = 1; n = 14; n = 1; n = 2; n = 10; n = 11; n = 12; n = 2; n = 2; n = 6; n = 5.

Epidemiology of Pediatric PMD

Among adult patients with neurological symptoms, up to 25% may not be fully explained by an organic neurological diagnosis. In adult movement disorder clinics the frequency of PMD ranges between 2 and 4%. Limited data are available on the population prevalence and incidence of psychogenic disorders in children. In a retrospective survey of 2280 children with neurological disorders, a psychogenic etiology was reported in 6.4%.19 Somatization presenting with neurological disorders, a psychogenic etiology was very common in children. However, conversion reactions seem to be more frequent in late childhood and adolescence.20,21 The most common symptoms are gait disturbances, respiratory symptoms, speech disturbances, and urinary retention. Our own 15 cases of pediatric PMD were seen in a specialist quaternary movement disorders clinic over the past 11 years, and this would suggest an estimated incidence of about 2%. This is in agreement with the frequency of pediatric PMD in a Spanish series of 666 children with movement disorders and resembles the frequency described in adults. However, it is difficult to try to extrapolate a population incidence of PMD in adults or children from the experience of specialist movement disorder clinics, and only population-based studies would be able to truly assess the incidence of PMD in children.

Clinical Characteristics and Diagnosis of Pediatric PMD

As with adult-onset PMD, the PMD in children are more frequent in females (our cases: F:M = 4:1; literature: F:M = 3.2:1). Only 13% of our cases had onset prior to the age of 10, with 7 as age at onset being the earliest. Similar to this, only 6 of 50 previously reported cases had onset prior to age of 10, the youngest being a 3-year-old boy.16 Clues from the history and the examination suggesting a movement disorder of psychogenic origin were nearly identical to those described in adults.23

The most common types of PMD among our cases were dystonia (47%), tremor (40%), and gait disorders (13%). Multiple hyperkinetic movement disorder phenomenologies were observed in many cases. This distribution resembles the data from the literature review dystonia (34%), gait disturbance (30%), tremor (26%), tics (6%), and myoclonus (4%). In adults, the most common PMD is tremor (55%), followed by dystonia (39%), myoclonus (13%), tics (6%), gait disorder (3%), and parkinsonism (2%), and combinations of different movement disorder types are common as well.24

In our series, the abnormal movement started in one limb in 10 cases. In nine of them, the dominant side was affected. This confirms the observation by Regan and LaBarbara, who additionally suggested that this may be the result of incomplete hemispheral lateralization in children.25 This is in contrast to adults where involvement of the nondominant limb seems to be characteristic.26

According to DSM IV criteria, the underlying psychiatric diagnosis was conversion disorder in the majority of our 15 cases (80%) and the reviewed cases (86%). Only 20% of our series and 9% of published cases were diagnosed with a somatization disorder, none of our cases and only 5% of published cases were diagnosed with a factitious disorder (deliberate production of symptoms for psychological gain). Malingering (deliberate production of symptoms for external/material gain) was not reported. This resembles the distribution of psychiatric diagnosis described in PMD in adults.1

PMD occurred as a single neurological diagnosis in the majority of cases. In 40% of our cases and 10% of the reviewed cases a coexisting organic disorder was reported. Among these, organic neurological diagnoses were the most common; however, an organic movement disorder was rarely reported (our cases: two with tics, one with dystonia; literature: one with dystonia2). This is perhaps surprisingly, since in adults the prevalence of coexistent psychogenic and organic movement disorders has been suggested to lie between 10 and 25%.4,11 Only 13% of our cases and 12% of the reviewed cases had a documented preexisting psychiatric comorbidity. In a study that evaluated the psychiatric comorbidity in 88 adult patients with PMD by structured clinical interviews, 38% had an axis I
diagnosis (e.g., major depression, anxiety).\textsuperscript{28} Despite the apparent lower rate of preexisting psychiatric comorbidity in the childhood-onset cases of PMD, standardized psychiatric assessment was not performed in all such cases, and, therefore, the rate of underlying psychiatric diagnosis may have been underreported.

Previously, published risk factors for development of PMD include history of sexual abuse or rape, previous surgery or other physical trauma, and major emotionally stressful life events, such as divorce or death of a family member.\textsuperscript{1,18,28} In our cohort as well as the 50 cases from literature, sexual or physical abuse was not reported, although a standardized assessment in this regard was not performed. A preceding minor injury was present in 40% of our cases and in 14% of the cases from the literature. Interestingly, out of all the 14 children with a preceding minor physical trauma, 12 developed dystonia which was fixed and accompanied by pain in the majority. Trauma preceding fixed dystonia is a well-known phenomenon in adults and has been reported to be as high as 63%.\textsuperscript{15} An emotionally stressful life event was frequently mentioned (overall in 39%).

Time from symptom onset to diagnosis of a PMD varied broadly (between 2 weeks and 5 years in our cases and between a few days to 21 years in the reviewed cases). With the exception of the children with psychogenic tremor, all children underwent extensive investigations to exclude a structural, biochemical, immunological, or genetic cause of their movement disorder. This suggests that clinicians are more confident in diagnosing psychogenic tremor on purely clinical grounds, in contrast to patients with dystonia, who often underwent very extensive testing for organic causes of the disorder. Although in most of the cases, clinical features strongly supported the diagnosis of a PMD, psychogenic dystonia was usually considered as a diagnosis of exclusion after very detailed investigation.

**Treatment and Outcome of Pediatric PMD**

Very little evidence exists regarding the best choice of treatment in PMD in adults.\textsuperscript{29,30} According to data from our childhood-onset cases and a review of the literature, a cognitive and behavioral approach to treatment and rehabilitation by a multidisciplinary team, which includes a clinical psychologist, physiotherapist, occupational therapist, neurologist, and psychiatrist, proves helpful in most of the children. In adult cases with recent or current anxiety or depression, treatment with antidepressants was suggested to be helpful.\textsuperscript{30} Among our 15 cases, 47% recovered fully, 33% improved substantially, and 20% remained chronically and severely disabled. Of 50 cases of pediatric PMD from the literature review, 74% recovered fully, 14% improved partially, 8% remained chronically disabled, and in 4% the outcome was not reported. Children with a brief duration of symptoms (<1 month) prior to diagnosis tended to recover spontaneously or with removal of a psychological stressor, suggestion, or psychotherapy. In general, children with tremor as predominant PMD tended to have a more favorable prognosis. In part, this might reflect the earlier diagnosis of PMD in patients with tremor compared with other movement disorders. It appears that the longer the disease duration prior to diagnosis, the less favorable is the prognosis. This may explain the worse prognosis of our cases in contrast to the ones described in the literature, as typically, only more difficult cases tend to be seen in a quaternary movement disorders clinic, and usually at a later stage of the disease. This also might explain why none of the cases seen by us recovered spontaneously or just with suggestion. However, even children with longstanding symptoms can improve with psychological support and the use of cognitive and behavioral techniques.

**CONCLUSIONS**

From our cases and the limited literature regarding PMD in childhood some conclusions can be drawn: (1) the same clinical clues suggest the presence of a PMD in children and adults, (2) the most common PMD in children are dystonia, tremor, and gait disorders, (3) unlike adults, in children with PMD the dominant side is more commonly involved, (4) psychogenic tremor is commonly diagnosed clinically, whereas other PMD (despite a typical clinical picture) often lead to extensive investigation, (5) children with a shorter disease duration prior to diagnosis (<1 month) have an excellent prognosis, and (6) those with a longer disease duration have a poorer prognosis, but treatment with cognitive and behavioral therapies and rehabilitation by a multidisciplinary team causes marked improvement and return to school and home life in the majority. On the basis of these data, we suggest that in children with suspected PMD, strenuous efforts should be made to reduce the time between symptom onset and diagnosis. Where there is diagnostic uncertainty, rapid referral to a movement disorder specialist for assessment and planning of appropriate investigation could assist with earlier diagnosis and introduction of appropriate treatment.
REFERENCES