ORIGINAL ARTICLE

Pediatric Functional Movement Disorders: Experience from a Tertiary Care Centre

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ABSTRACT: ***Objectives:*** Functional movement disorders (FMDs) pose significant diagnostic and management challenges. We aimed to study the socioeconomic and cultural factors, underlying psychopathology and the phenomenology of FMDs in children. ***Methods:*** The study is a retrospective chart review of 39 children (16 girls and 23 boys) who attended our neurology OPD and the movement disorders clinic at the National Institute of Mental Health and Neurosciences (NIMHANS) between January 2011 and May 2020. The diagnosis of FMD was based on Fahn and Williams criteria and the patients were either diagnosed as "documented" or "clinically established". All the relevant demographic data including the ethnicity, socioeconomic and cultural background, examination findings, electrophysiological, and other investigations were retrieved from the medical records. ***Results:*** The mean age at onset was 12.69 ± 3.13 years. Majority of the children were from urban regions (56.41%) and belonging to low socioeconomic status (46.15%). Thirty (76.92%) were found to have a precipitating factor. Myoclonus was the most common phenomenology observed in these patients (30.76%), followed by tremor (20.51%), dystonia (17.94%), and gait abnormality (7.69%). Chorea (5.12%) and tics (2.56%) were uncommon. Tremor (37.5%) and dystonia (18.75%) were more common in girls, whereas myoclonus (39.13%) was more common in boys. ***Conclusions:*** The symptoms of FMD have great impact on the mental health, social, and academic functioning of children. It is important to identify the precipitating factors and associated psychiatric comorbidities in these children as prompt alleviation of these factors by engaging parents and the child psychiatrist will yield better outcomes.

RÉSUMÉ : Les troubles fonctionnels du mouvement chez les enfants dans le contexte d'un établissement de soins tertiaires. ***Objectifs*** **:** Les troubles fonctionnels du mouvement (TFM) soulèvent des enjeux importants en matière de diagnostic et de prise en charge des patients. C'est ainsi que nous avons voulu nous pencher sur les facteurs socioéconomiques et culturels qui sous-tendent la psychopathologie et la phénoménologie des TFM chez les enfants. ***Méthodes*** **:** Cette étude est fondée sur une analyse rétrospective des dossiers de 39 enfants (16 filles et 23 garçons) qui ont fréquenté, entre janvier 2011 et mai 2020, notre service de consultations externes en neurologie et la clinique des troubles du mouvement du *National Institute of Mental Health and Neurosciences* (NIMHANS). Les diagnostics de TFM ont reposé sur les critères de Fahn et Williams. À noter que ces patients ont été divisés en deux groupes en ce qui regarde leur diagnostic : ceux étant probablement atteints (on dira « documentés ») et ceux dont le diagnostic a été établi sur le plan clinique. Toutes leurs données démographiques pertinentes, ce qui inclut leur appartenance ethnique et leur contexte socioéconomique, de même que leurs résultats d'examens, notamment d'un point de vue électro-physiologique, ont été obtenus à partir de leurs dossiers médicaux. ***Résultats*** **:** L'âge moyen d'apparition des premiers symptômes de TFM était de 12,69 ± 3,13 ans. La majorité des enfants provenaient de régions urbaines (56,41 %) et étaient issus de milieux socioéconomiques défavorisés (46,15 %). Sur ces 39 enfants, on a noté que 30 d'entre eux (76,92 %) donnaient à voir un facteur précipitant. La myoclonie est la manifestation clinique qui a été la plus observée chez ces patients (30,76 %) ; ont suivi les tremblements (20,51 %), la dystonie (17,94 %) et les anomalies de la démarche (7,69 %). La chorée (5,12 %) et des tics nerveux (2,56 %) se sont par ailleurs avérés peu fréquents. Enfin, les tremblements (37,5 %) et la dystonie (18,75 %) étaient plus fréquents chez les filles tandis que la myoclonie (39,13 %) l'était chez les garçons. ***Conclusions*** **:** Les symptômes de TFM ont un impact notable sur la santé mentale et sur le fonctionnement social et scolaire des enfants. Il importe donc d'identifier les facteurs précipitants et les comorbidités psychiatriques associées chez ces enfants car une atténuation rapide de leurs conséquences, en impliquant les parents et les pédopsychiatres, débouchera sur une meilleure évolution de leur état de santé.

Keywords: Functional, Movement disorder, Tremor, Dystonia, Myoclonus, Precipitating factors

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Introduction

Functional movement disorders (FMDs) pose significant diagnostic and management challenges for the neurologist.[1] These disorders (FMDs) are well characterized in adults, but childhood-onset FMDs have not been extensively studied.[2] FMDs are also more common in adults and uncommon or rare

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in children.[3,4] FMDs account for about 1.5% of all the patients attending neurology clinics and 17% of all functional neurological disorders.[5] In the movement disorders clinic, the estimated prevalence is about 2%–10%.[6] About 10% of them are found to have associated organic disorder.[7] However, the exact prevalence is difficult to estimate due to the case definition, referral bias, difficulty in differentiating from the organic disorders, and low index of suspicion by the treating physician.

These disorders do not have any structural or biochemical abnormality, but are believed to be due to underlying psychological or psychiatric illness.[8,9] They are thus a part of functional neurological disorders. In recent years, there are controversies regarding the terminologies "psychogenic" or "functional" for these disorders.[10] The diagnostic criteria were initially laid by Fahn and Williams[11] in patients with psychogenic dystonia that was later modified to include other FMDs.[9,11]

Given the paucity of literature on these disorders in children, the study of these disorders in children becomes essential as it affects the mental and social well-being of the child. In addition, it is associated with school absenteeism, poor scholastic performance, lack of self-confidence, and parental anxiety. Hence, we aimed to study the socioeconomic and cultural factors, underlying psychopathology and the phenomenology of FMDs in children.

Methods

The present study was a retrospective chart review of 39 children (aged ≤ 18 years) who attended our neurology OPD and the movement disorders clinic between January 2011 and May 2020. Among them, the clinical profile of 22 children has been published earlier.[3] The study was approved by the institute's ethics committee. The diagnosis of FMD was based on the criteria described by Fahn and Williams and Gupta and Lang.[9,11] All these children were examined by the senior movement disorder specialist (PKP and RY) with clinical opinion from the child psychiatrist (SS). All the relevant demographic data including the ethnicity, socioeconomic and cultural background were documented. Detailed history, examination findings, electrophysiological, and other investigations were also noted. The clinical videos were also reviewed. Most of these children had undergone electrophysiological evaluations such as surface electromyogram (EMG), long-loop reflexes, jerk-locked back averaging, and Bereitschaftspotential (BP) depending on the underlying phenomenology. The psychiatric notes were also reviewed and discussed with the child psychiatrist (SS). The treatment details and the outcome were also recorded.

Statistical Analysis

The statistical analysis was performed using the R software. All the variables were expressed as mean ± standard deviation, percentage, and range.

Results

Demographic and Clinical Characteristics (Table 1)

Thirty-nine children (16 girls and 23 boys) were evaluated during the 9-year period (2011–2020). The mean age at onset was 12.69 ± 3.13 years with a median duration of illness being 150 days (range 2 days– 6 years). About 56.4% of children were from urban regions. Majority (66.7%) of the children belonged to low-to-middle socioeconomic strata (low – 46.15%; middle – 20.51%). The onset of the illness was acute in 69.23% and chronic presentation in 25.64%. Antecedent illness was observed in 14 children with joint pains being the most common (28.57%). Other antecedent illnesses that were noted included fever (21.42%), loss of consciousness (14.28%), headache (7.14%), speech disturbance (7.14%), eye pain (7.14%), and abdominal pain (7.14%). Thirty children were found to have a precipitating factor in the form of stressors at school (36.66%), combined school and family stressors (26.66%), family stressors (23.33%), and societal pressures (13.33%).

FMD Phenomenology (Table 2)

Myoclonus was the most common phenomenology observed in these patients (30.76%), followed by tremor (20.51%), dystonia (17.94%), and gait abnormality (7.69%). Chorea (5.12%) and tics (2.56%) were uncommon. Tremor (37.5%) and dystonia (18.75%) were more common in girls, whereas myoclonus was more common in boys (30.76%). Gait abnormality was seen exclusively in boys (13.04%). Multiple FMD phenomenologies were seen in two girls and one boy. One girl presented with functional vocal tics and one boy with functional dysphonia.

Course and Outcome

Electrophysiological testing was performed in 17 children (43.58%) and FMD was confirmed in all of them. In patients with functional tremors, surface EMG showed variability, distractibility (Figure 1), and entrainability (Figure 2). BP was demonstrated in some patients with myoclonus (Figure 3). Placebo response was observed in 30.6% of children. Nineteen children (48.71%) required hospital admission. Twenty-two (56.41%) showed complete resolution of the symptoms and in 11.1%, there was a partial improvement. All 39 children in our study were referred to child psychiatry specialists for detailed evaluation and majority ($n = 37$, 94.9%) of them were diagnosed with a dissociative movement disorder. Associated psychiatric comorbidity was observed in 12.8% in the form of obsessive-compulsive disorder, depression, generalized anxiety disorder, schizophrenia, and psychosis.

Discussion

FMDs are heterogeneous disturbances of motor function that are not explained by organic conditions and may occur in association with underlying psychiatric disease.[12] FMD poses a great challenge for clinicians in diagnosis and management.

In our study, 39 children with documented and clinically established FMD were evaluated. The most common type of FMD observed was myoclonus, tremor, and dystonia. The exact prevalence or frequency of pediatric FMD varies between 2% and 3% of children attending the movement disorders clinic.[2,12,13] In a study involving 2280 children with neurological disorders, psychogenic etiology was observed in 6.4%.[14]

In our study, FMD was more common in boys, which is similar to the previous two studies from India.[3,15] However, some previous studies have shown the higher prevalence in girls.[2,12,16,17] In another study from our institute, about 48% of patients with the then-existing diagnostic category of hysteria were girls.[18] The reason for predominance of FMD in boys is not clear, but it may be due to sociocultural practices prevailing in the Indian community wherein medical attention is sought more often and earlier for

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Table 1: Demographic and clinical characteristics of the children

Characteristic	Total children ($n = 39$)	Girls ($n = 16$)	Boys ($n = 23$)
Age (years) (mean ± SD)	13.64 ± 3.08	13.69 ± 3.14	13.64 ± 3.06
Age at onset (years) (mean ± SD)	12.69 ± 3.13	12.66 ± 3.19	12.69 ± 3.13
Rural/urban			
Rural	17 (43.59%)	7 (43.75%)	10 (43.47%)
Urban	22 (56.41%)	9 (56.25%)	13 (56.53%)
Socioeconomic status			
Low	18 (46.15%)	8 (50%)	10 (43.47%)
Middle	8 (20.51%)	2 (12.5%)	6 (26.08%)
High	13 (33.33%)	6 (37.5%)	7 (30.43%)
Onset of illness			
Abrupt	27 (69.23%)	10 (62.5%)	17 (73.91%)
Subacute	2 (5.12%)	1 (6.25%)	1 (4.34%)
Chronic	10 (25.64%)	5 (31.25%)	5 (21.73%)
Antecedent illness	14 (35.9%)	4 (25%)	10 (43.48%)
Joint pain	4 (28.57%)	0	4 (40%)
Headache	1 (7.14%)	1 (25%)	0
Fever	3 (21.42%)	2 (50%)	1 (10%)
Loss of consciousness	2 (14.28%)	1 (25%)	1 (10%)
Speech disturbance	1 (7.14%)	0	1 (10%)
Eye pain	1 (7.14%)	0	1 (10%)
Abdominal pain	1 (7.14%)	0	1 (10%)
Trunk bending	1 (7.14%)	0	1 (10%)
Precipitating factors	30 (76.92%)	13 (81.25%)	17 (73.91%)
Stressors at school	11 (36.66%)	4 (30.77%)	7 (41.17%)
Stressors at family	7 (23.33%)	3 (23.07%)	4 (23.53%)
Societal stressors	4 (13.33%)	1 (7.7%)	3 (17.65%)
Combined school and family stressors	8 (26.66%)	5 (38.46%)	3 (17.65%)

Table 2: Phenomenology of the movement disorders observed in children

Phenomenology	Total children ($n = 39$)	Girls ($n = 16$)	Boys ($n = 23$)
Tremor	8 (20.51%)	6 (37.5%)	2 (8.69%)
Dystonia	7 (17.94%)	3 (18.75%)	4 (17.39%)
Myoclonus	12 (30.76%)	3 (18.75%)	9 (39.13%)
Gait	3 (7.69%)	0	3 (13.04%)
Tics (motor)	1 (2.56%)	0	1 (4.34%)
Chorea	2 (5.12%)	1 (6.25%)	1 (4.34%)
Facial spasm	1 (2.56%)	0	1 (4.34%)
Combined gait and dystonia	1 (2.56%)	1 (6.25%)	0
Combined tremor and dystonia	1 (2.56%)	0	1 (4.34%)
Combined tremor and myoclonus	1 (2.56%)	1 (6.25%)	0
Vocal tics	1 (2.56%)	1 (6.25%)	0
Dysphonia	1 (2.56%)	0	1 (4.34%)

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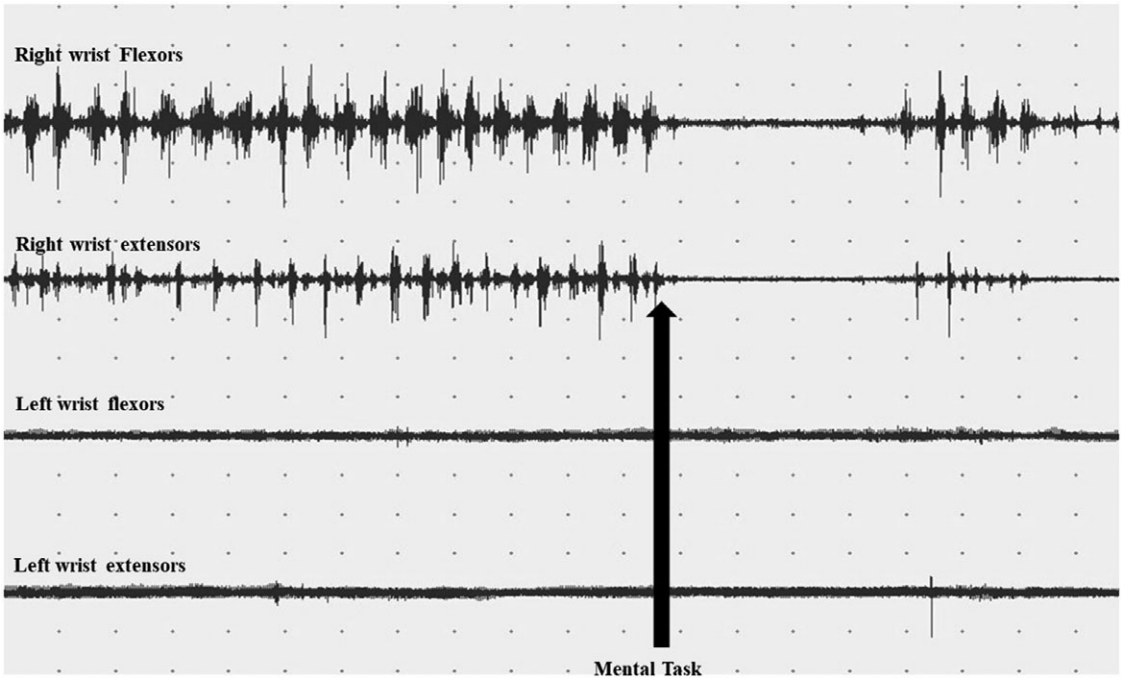

Figure 1: *Distractibility in a patient with functional tremor. During the mental task, there is abrupt cessation of tremor in the right upper limb.*

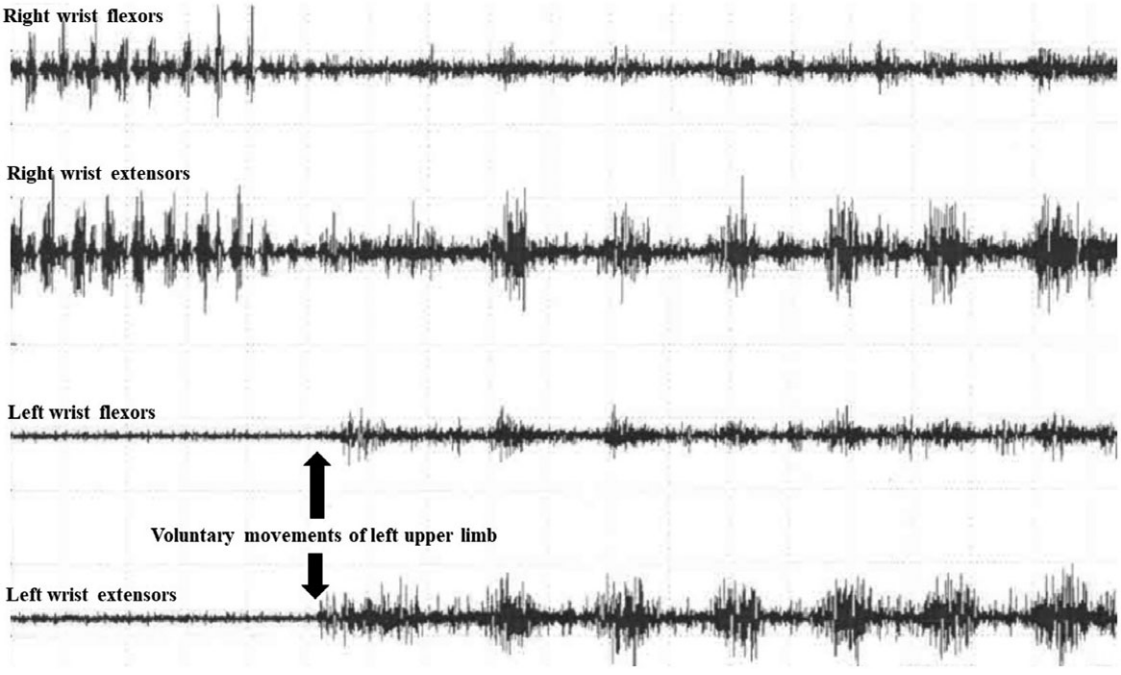

Figure 2: *Entrainability in a patient with functional tremor. During voluntary movements of the left upper limb, there is abrupt cessation and change in the tremor frequency and amplitude in the right upper limb.*

the boys than girls. Indian society is mainly patriarchal.[19] There is a preference for a son in most of the Indian families and there is discrimination against girls especially in providing education and medical care.[20]

In our study, majority of the children belonged to low-to-middle socioeconomic strata. In India, patients from low-to-middle socioeconomic populations more often visit government hospitals, like ours. However, to the best of our knowledge, there is no published literature from India on the prevalence of pediatric FMD based on socioeconomic status.

Abrupt onset of symptoms is one indicator toward FMD, which was seen in 69.2% of our cohort. In a study of 54 children,

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Table 3: Spectrum of PMD phenomenology in other studies

Author	Sample size	Study methods	Results
Ferrara J et al.[2]	Fifty-four children	Review of the medical records of children who were diagnosed with FMDs since 1988.	The mean age at symptom onset was 14.2 ± 2.11 years (range 7.6–17.7). Two-thirds of children exhibited multiple FMD phenotypes, the most common being tremor (65%), followed by dystonia (43%), myoclonus (37%), and gait (22%).
Ahmed MAS et al.[16]	Thirty-four children (23 organic movement disorders, 5 documented FMDs, and 6 probable or possible FMDs)	Prospective study over a period of 2 years (August 2004–August 2006). All subjects were clinically evaluated and followed up for a period ranging from 1 to 3 years.	Tics in six boys, tremors in four (three girls), and myoclonus in one girl.
Schwingenschuh P et al.[12]	Fifteen children	Retrospective review of the medical records of all children (aged ≤ 18 years) who were diagnosed with FMD between 1996 and 2007 in the Pediatric Movement Disorders Clinic of Great Ormond Street Hospital, London (under the care of late Robert Surtees).	The mean age at onset was 12.3 ± 2.6 years (range 7–16 years). 80% were female (F:M=4:1). The most common movement disorder was dystonia ($n = 7$), followed by tremor ($n = 6$) and two with gait disturbance. In six children (40%), FMD had a single phenomenology, while the remaining nine children exhibited multiple motor phenotypes.
Faust J et al.[24]	Thirteen children (10 girls)	Retrospective review of all patients is seen in the pediatric neurology movement disorders clinic. This was conducted using a clinic database, with chart review and review of video recordings, were available.	Most common phenomenology was dystonia in five children, followed by tremor in three, chorea in three, and myoclonus in three children.
Canavese C et al.[17]	Fourteen children (6 boys and 8 girls)	Retrospectively reviewed medical records and video of patients <18 years diagnosed with FMD between 2007 and 2010.	Most frequent movement disorders were tremor (36%) and dystonia (29%).
Kamble et al.[3]	Seventy-three PMD (22 children)	Retrospective chart review of patients with FMD was evaluated between 2000 and 2013.	The mean age of onset was 12.1 ± 3.4 (8–18) years. Most frequent FMD was myoclonus (36.4%) followed by tremor + myoclonus (13.6%), tremor (9%). and tremor + dystonia (9%).
Pandey S et al.[15]	Twenty-five children (12 boys and 13 girls)	Detailed report of patients with FMD who attended the movement disorders clinic over the past 2 years.	The mean age was 14.12 ± 2.48 years. Tremor 44%, dystonia 16%, gait abnormality 16%, tics 12%, myoclonus 0.08%, and abdominal dyskinesia 0.04%.
Present study	Thirty-nine children (23 boys and 16 girls)	Retrospective chart review of patients with FMD was evaluated between 2011 and 2020.	The mean age of onset was 12.69 ± 3.13 (5–18) years. Most frequent FMD was myoclonus (30.76%) followed by tremor (20.51%), dystonia (17.94%), gait abnormality (7.69%), and chorea (5.12%).

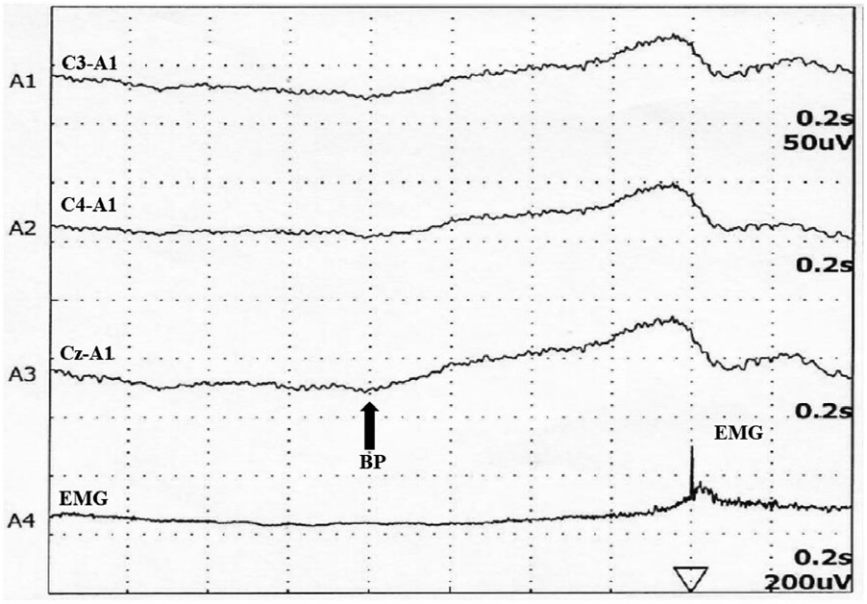

Figure 3: *Bereitschaftspotential (BP) in a child with functional myoclonus. BP is seen about 800 msec before the electromyogram (EMG) activity.*

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approximately 90% of children presented with an abrupt onset.[2] Many other studies have also reported a preponderance of abrupt onset of illness.[17] In addition, antecedent illness is commonly seen in these patients. In our study, nearly a third of the children had an antecedent illness which is similar to the other studies.[3,17]

History of precipitating factors needs to be enquired in patients suspected of FMD. Most of the studies have reported precipitating factors ranging from 47.9% to 83.7% in adults.[21,22] The various precipitating factors identified in these studies include physical injury, psychological insult, flu-like illness, death of a relative, poverty, family, or social stress.[21–23]

In a study involving 38 children, significant stressors were identified in 27 that included academic difficulties in school, punitive parent, parental discord, financial difficulties, increasing workload, and sibling rivalry.[18] Traumatic experience (physical or psychological) and emotional factors in the childhood have a significant impact on the pathogenesis of FMD.

Myoclonus followed by tremor and dystonia were the common phenomenologies observed in our study. However, tremor[2,17,18] and dystonia[12,24] have been the most common FMD in other studies.

Detailed psychiatry evaluation is necessary for diagnosis and treatment of FMD. The various psychiatric comorbidities that have been associated with FMD include obsessive-compulsive disorders, depression, and anxiety.[25]

Clinically, it is challenging to diagnose FMD. However, some cues that may be useful for diagnosis can be categorized as historical (abrupt onset, static course, psychiatric comorbidity, precipitating factors, presence of multiple somatizations, pending litigation, etc.), clinical (incongruence, inconsistency, distractibility, variability, entrainability, fluctuating course, suggestibility/modulation, episodic occurrence with complete or incomplete remissions, spontaneous remissions, and additional movement disorders), and treatment related (response to psychotherapy, placebo).[22] Electrophysiological methods such as surface EMG recording for tremors and BP in myoclonus may be helpful in determining the functional basis for the movement disorders.[3]

Recent neuroimaging studies have implicated abnormal emotional processing, sense of agency, and top-down regulation from frontal areas as important pathophysiological components in FMD.[25] Abnormal regional brain activations and functional connectivity have been demonstrated using functional MRI and positron emission tomography (PET).[26] Abnormal connectivity is found in right caudate, amygdala, prefrontal, and sensorimotor regions in patients with FMD with over 68% sensitivity and specificity.[27] In addition to the abnormal activation of supplementary motor area (SMA), patients with functional tremors and dystonia have abnormal activation in the right middle temporal gyrus or right temporoparietal junction.[28] The cingulate cortex has been implicated in self-awareness, self-monitoring, and active motor inhibition.[29] In patients with functional tremors and dystonia, there is abnormal activation of the cingulate cortex.[30] In future, these advanced imaging techniques may be useful in the diagnosis of FMD. Currently, these studies have been done only in adults. There are no such studies in pediatric FMD.

In our study, 56.4% of children had complete resolution of symptoms, and in 11.1%, there was a partial improvement and 13.9% showed no improvement at all, which may be due to non-acceptance of functional nature of the disease by parents/patients. There was no child with a worsening of symptoms. Studies have reported response rates ranging from 5% to 57%.[23,30–33] Similar good outcome has been observed in previous studies from India.[15] In another study, the outcome was not described due to lack of follow-up which is a problem with patients with FMD.[2] In a study by Schwingenschuh et al., 47% improved completely, 33% improved substantially, and 20% remained chronically and severely disabled.[12] A summary of the previous and present studies is shown in Table 3.

Conclusions

In our study, we observed that FMD was more common in boys and myoclonus as the most common phenomenology. This is in sharp contrast to previous studies. This may be due to gender bias as well as social stigma in our country. The symptoms of FMD have a great impact on the mental health, social, and academic functioning of children. It is important to identify precipitating factors and associated psychiatric comorbidity in these children as prompt alleviation of these factors by engaging parents and the child psychiatrist will yield better outcomes. Electrophysiology and placebo effect may serve as useful supplementary tools for diagnosing FMD.

Conflict of Interest

None of the authors have any financial disclosure to make or have any conflict of interest.

Statement of Authorship

KR: Data collection, manuscript writing, study design, and data review.

NK: Data collection, manuscript writing, study design, and data review.

RY: Manuscript review.

AB: Data collection, statistical analysis.

VVH: Manuscript review.

MN: Manuscript review.

SS: Study design, supervision, data review, and manuscript review.

PKP: Conception of the study, study design, supervision, data review, and manuscript review.

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