

Functional neurological movements in children: Management with a psychological approach

Sally Robinson^{a, b}, Raj Seraya Bhatia^a, Tamsin Owen^a, Kate Golding^b, Osman Malik^a, Tammy Hedderly^{a, c, *}

^a Tic and Neurodevelopmental Movements Service (TANDeM), Evelina Children's Hospital, Guys and St Thomas' NHS Foundation Trust, London, UK

^b Paediatric Neuropsychology Service, St Georges University Hospitals, London, UK

^c Paediatric Neurosciences, Kings College London, UK

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ABSTRACT

Aim: To develop a cognitive behavioural treatment (CBT) approach that included novel attention training components to support symptom management in children with a primary diagnosis of Functional Movement Disorder (FMD).

Method: Eighteen children (9 male and 9 female) with a mean age of 13 years (sd = 2.46, range 10–18 years) were assessed and completed CBT with novel attention training components. Treatment outcomes were measured using the Child Global Assessment Scale (CGAS) which was administered at baseline and post-treatment.

Results: Scores on the CGAS improved significantly post-treatment ($p < 0.001$) with all participants showing significant change in functioning on the basis of the Reliable Change Index (RCI), with clinically significant change across classification boundaries.

Interpretation: This case series provides support for the use of CBT with attention training components for the management of FMD. Larger trials are necessary to identify which individual treatment components are most effective and to better understand and quantify response to treatment. Future clinical treatment studies would benefit from the inclusion of objective measures of interoception and attentional focus.

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1. Introduction

Functional movement disorders (FMD) are reported by roughly a third of new neurology outpatients and around 15% go on to receive a primary functional diagnosis (e.g. non-epileptic seizures, functional weakness) [1,2]. Studies in children have reached broadly similar conclusions regarding epidemiology of FMD; age of onset is usually around early adolescence (approximately 11–14 years) and more commonly seen in girls with a surveillance study indicating 3:1 female to male ratio [3–6]. Within paediatrics, disturbances of motor function are the most common functional symptom (motor weakness, 63%; abnormal movements, 43%),

followed by non-epileptic seizures (40%) and sensory disturbances (32%), with 69% of patients reporting multiple symptoms and 79% requiring inpatient admissions [3]. There are increasing diagnoses in children with autism spectrum disorder, suggesting that there are likely to be some neurogenetic influences [7].

Diagnostic criteria and assessment of functional movement disorders (FMD) remains contentious, but all broadly encompass the description posited by Edwards and Bhatia (2012) - “A movement disorder that is significantly altered by distraction or non-physiological manoeuvres (including dramatic placebo response) and that is clinically incongruent with movement disorders known to be caused by neurological disease” [8]. This description highlights features that facilitate a ‘positive diagnosis’, rather than one based solely upon the exclusion of alternative medical explanations [9]. Patients often experience prolonged delays before a diagnosis is reached [4–6], with many patients diagnosed and then discharged due to a lack of appropriate services and access to therapeutic

* Corresponding author. TANDeM Service, Children's Neurosciences, Evelina London Children's Hospital, Guys and St Thomas Trust, Westminster Bridge Rd, London, SE1 7EH, UK.

E-mail address: tammy.hedderly@gstt.nhs.uk (T. Hedderly).

support [10].

Onset is typically abrupt and may be preceded by a physical or psychological difficulty [11], although evidence of psychological trauma is no longer required within the DSM-V criteria [12]. Common precipitating factors for functional symptoms in childhood include school related difficulties, interpersonal family conflicts, stressful life events and minor injuries [6,13], with consequences including emotional disturbances, lost educational opportunities, social isolation and increased family burden [14,15]. A triggering event is not always identified and importantly its presence is not necessary for diagnosis. As yet, no consensus has been reached about what causes or maintains FMD, though a biopsychosocial model is implicated [16].

Neuroimaging studies show significant differences in brain activation (especially the temporo-parietal junction), with atypical activation and connectivity between regions involved in self-agency [17–19]. Overactivity of the limbic system, greater motor preparation, increased connectivity between the limbic and motor systems and atypical somatomotor networks have also been reported [19–23]. These studies have contributed to theories proposing that FMD are linked with changes in top-down and bottom-up control mechanisms, with disruption to the monitoring of sensory feedback, motor control and attentional processes [24–27]. We do not therefore recommend ever using the term non-organic when explaining FMD to children or families.

From a clinical perspective, for adults with functional tremors, significant differences have been reported in the amount of visual attention directed towards the affected limb during examination [28], with variability or even cessation of functional tremors when attention is directed elsewhere [8]. Subjective experiences of FMD are influenced by attention, with patients self-reporting functional tremors for 80–90% of the day, despite tremors only presenting for 30 min [29]. Changes in interoceptive processes have also been reported; with poorer performance when attending to internal physiological information (such as a heartbeat) and emotional states [30]. These studies highlight the impact of attention on symptoms and suggest that treatments aimed at targeting attentional networks may help improve symptoms and outcome in FMD.

Along with physiological characteristics linked with higher rates of FMD, there is also some research in psychological characteristics in FMD such as Alexithymia (lack of ability to feel and describe emotions or ascribe emotional states to self). In one study, Alexithymia was present in 34.5% of patients with Functional Motor Symptoms, as compared with 9.1% with neurological movement disorders and 5.9% of the healthy volunteers, thus was significantly higher in the FMS group ($p < 0.001$) even after controlling for the severity of the symptoms of depression [31].

There is a limited evidence base for the use of psychological treatments for children with FMD, though a multidisciplinary approach is advocated, with inpatient units adopting the use of physiotherapy to support movement rehabilitation and psychological therapy to identify stressors and teach stress management strategies [6,15,22,32,33]. Recently, McFarlane et al. (2019) described a case series of young people who received a goal-based cognitive behavioural treatment for functional neurological symptoms (predominantly non-epileptic seizures, but also motor and sensory symptoms) [34]. There is also an emerging evidence base for the effectiveness of attention training in reducing anxiety for highly self-focused individuals [35]. However, to date, there are no studies reported in the literature that incorporate attention training components for the management of FMD.

1.1. Aims

The current study aimed to develop a cognitive behavioural

treatment approach that included attention training components to support symptom management in children with a primary diagnosis of FMD.

2. Methods

2.1. Participants

The children reported in this case series represent referrals to the Tic and Neurodevelopmental Movement (TANDeM) service at Evelina London Children's Hospital and Paediatric Neuropsychology Service at St Georges University Hospital, between 2014 and 2018. At referral the diagnosis of FMD had been established in some cases, but not others. In all cases, the diagnosis was confirmed clinically by the teams' neurologist, following investigations, as required. Children reported in this case series all presented with a movement difficulty as their primary presenting concern. There were several children whom were identified to have co-occurring neurological conditions (e.g. Tourette syndrome [TS]) and/or neurodevelopmental disorders (e.g. autism spectrum disorders [ASD]). The case series consists of 18 children/young people. The characteristics of all participants are reported in Table 1.

2.2. Assessment and measures

Patients underwent a multidisciplinary diagnostic assessment, led by a Consultant Neurologist and Consultant Psychiatrist, with Clinical Psychology. This included detailed history-taking and interviewing with the parent/carer(s) and child/young person both together and separately, with assessments typically lasting two to 3 h in duration. All assessments included a full developmental and medical history and a neurological examination, with the addition of a mental health assessment and a detailed review of medical notes and previous mental health reports. In some cases, we also used a 'complex case template' (Appendix 1), which we designed for use in our service to gather information in advance of the appointment from the GP about previous involvement from services and social services records. This has been especially useful if there are any concerns about a probable health anxiety or in cases of fabricated and induced illness in the family.

There is a lack of appropriate clinical tools for assessing FMD in children and adults, with standard mental health measures reported to have poor validity for patients with functional movement symptoms [34,36]. As such, assessment of functional symptoms and co-morbid mental health concerns were made based on the clinical assessment using the Children's Global Assessment Scale (CGAS) [37]. The CGAS is a global, clinical-rated measure used to assess social, emotional and behavioural functioning from 0 (very poor) to 100 (very high functioning). CGAS ratings were made by at least two clinicians, who were both present at the initial assessment and following clinical discussion at the end of treatment.

The CGAS has been reported to have good concurrent and discriminant validity and reliability between raters, with adequate test-retest reliability [37]. It has also been reported to be sensitive to change for patients with functional movement symptoms [34]. The reliable change index (RCI) represents the change score between pre- and post-treatment scores sufficient to reject the null hypothesis that the change is due only to measurement error $p < 0.05$. The CGAS has an RCI score of 10.73. Any observed individual change must exceed the RCI for it to be judged statistically significant [38], where clinically significant change also required a shift across a clinical cutoff boundary.

Table 1
Participant characteristics and CGAS scores.

Patient	Age at onset	Age at tx	Gender	Functional and Clinical Symptoms	Clinical Input	Outcome	CGAS pre	CGAS post
1	12	14	Male	Functional weakness with non-walking and collapse, headaches, blurring of vision, stuttering, salivating, non-school attendance, social anxiety	14 psychological therapy sessions Neuropsychological assessment	Functional symptoms resolved, school support provided for specific learning difficulties	46	80
2	14	16	Female	Fine resting tremor of both hands, leg tremor, head tremor, intermittent loss of movement in legs, social anxiety	17 psychological therapy sessions +5 follow-up sessions Psychiatric review with medication (Sertraline) MRI - small benign lesion right frontal lobe	Functional symptoms resolved. School support provided for anxiety. Re-presented with low mood and referred to CAMHS. Referred to benign brain lesion clinic for monitoring	42	66
3	6	10	Male	Tics, functional movements with shaking of right arm, leg and bottom, loss of walking	2 psychological therapy sessions Psychiatric review	Functional symptoms resolved, tics well-managed	50	75
4	9	10	Male	Functional loss of lower limbs, ADHD, ASD, non-school attendance	6 psychological therapy sessions Psychiatric review Child protection meetings	Functional symptoms resolved, reintegrated in school with EHCP to manage ASD and ADHD	42	71
5	13	14	Female	Functional non-walking, chronic pain, non-school attendance	17 psychological therapy sessions Neuropsychological assessment	Functional symptoms resolved, school support provided for specific learning difficulties, part time job	44	84
6	15	15	Female	Functional right-hand stiffness, fixed position of hand/wrist	2 psychological therapy sessions	Functional symptoms resolved, with occasional cramps when writing	62	88
7	10	15	Male	Tics, functional head throwing	6 psychological therapy sessions	Functional symptoms resolved, school support provided to manage tics and anxiety.	58	80
8	10	10	Female	Functional loss of movement in leg, tics, functional tic-like attacks, obsessive compulsive behaviours, ASD	11 psychological therapy sessions	Functional symptoms resolved	35	60
9	11	11	Male	Functional loss of movements lower limbs, PANDAS	11 psychological therapy sessions	Functional symptoms resolved	35	75
10	8	16	Female	Psychogenic non-epileptic seizures, anxiety	22 psychological therapy sessions Psychiatric review with medication (sertraline)	Functional symptoms resolved	55	90
11	10	13	Female	Clawed hands and feet	7 psychological therapy sessions Neuropsychological assessment (+local physiotherapy)	Functional symptoms resolved, school support for missed learning and to manage anxiety. Represented with low mood and referred to CAMHS	40	72
12	11	16	Female	Tremor of hands/arms	15 psychological therapy sessions Neuropsychological assessment	Functional symptoms resolved with tremor only noticeable when anxious	45	70
13	15	18	Female	Tics, functional tic-like attacks, loss of movement in legs, complex motor stereotypies, social anxiety, non-school attendance	12 psychological therapy sessions	Reduction in functional tic-like attacks and increased control of motor stereotypies	36	60
14	15	16	Female	Tics, functional tic-like attacks, social anxiety	8 psychological therapy sessions Psychiatric review with medication (propanalol)	Functional tic-like resolved. Referred to local CAMHS for anxiety management	45	60
15	10	15	Male	Tics, functional tic-like attacks, impulse control disorders, OCD, ASD, non-school attendance	10 psychological therapy sessions	Functional tic-like attacks resolved, tics well-managed, improvement in OCD	31	78
16	12	13	Male	Tics, functional tic-like attacks, school non-attendance	6 psychological therapy sessions Parents attended tic psychoeducation groups	Functional tic-like attacks resolved, reintegrated at school	42	76
17	15	15	Male	Tics, functional tic-like attacks	1 psychological therapy sessions Psychiatric review	Functional tic-like attacks resolved	51	71
18	10	11	Male	Tics, functional tic-like attacks, social anxiety, school non-attendance	7 psychological therapy sessions	Functional tic-like attacks resolved	40	71

tx = treatment; MRI = magnetic resonance imaging; CAMHS = Child and Adolescent Mental Health Service; ADHD = Attention Deficit Hyperactivity Disorder; ASD = Autism Spectrum Disorder; EHCP = Education and Health Care Plan; PANDAS = Paediatric Autoimmune Neuropsychiatric Disorders Associated with Streptococcal Infections; OCD = Obsessive Compulsive Disorder.

2.3. Intervention (see Fig. 1 for details)

The children/young people in this case series received an individually-tailored psychological intervention for management of FMD and co-occurring mental health concerns. The treatment was delivered by a Paediatric Clinical Neuropsychologist or Clinical Psychologist, with nurse liaison provided to the school and family as needed (e.g. sharing care plans, supporting school staff to understand FMD). Sessions typically took the form of joint family sessions for psychoeducation and management strategies for FMD, followed by individual treatment sessions with the child/young

person that the parent/carer(s) joined for the first 5–10 min and last 5–10 min. Sessions were typically 60 min long. On average children received 9 sessions of psychological therapy for management of FMD and co-occurring concerns.

The initial phase of treatment was the multidisciplinary team (MDT) assessment which in line with guidance in the adult literature included therapeutic history-taking, showing the child and family the 'positive signs' of FMD and providing an explanation for symptoms [9]. As part of this assessment, the child's symptoms were formulated within a biopsychosocial framework that took into consideration predisposing (e.g. genetic, personality traits,

coping skills, family functioning, modelling), precipitating (e.g. injury, disease, mental health, cognitive profile, life changes, social stressors), perpetuating (e.g. muscle tension, pain/fatigue, illness beliefs, symptom checking, avoidance, aids, reorganisation around the illness, education/welfare system) and protective (e.g. help seeking, family/educational support) factors. This formulation was shared with the child and family at the initial assessment and revisited throughout treatment, with refinement of the formulation as new information emerged. Treatment was only provided if the family were accepting of the diagnosis and following completion of all medical investigations.

The psychological treatment program included typical cognitive behavioural therapy components, but with an added emphasis on the role of attentional focus to the onset and maintenance of functional movement symptoms (see Fig. 1). All patients and their families received psychoeducation about FMD and attention as a cognitive process (e.g. selective attention, sustained attention, divided attention), with additional psychoeducation around anxiety and other mental or physical health conditions as indicated. Behavioural experiments to help demonstrate the effects of attentional focus on physiological sensations and functional symptoms were included as part of the psychoeducation session. Attention training exercises (based on the metacognitive therapy literature) were also used with some patients to encourage increased external attention focusing, with sensory grounding techniques taught to help reduce attentional focus on internal bodily sensations when anxious so that they could then engage with cognitive strategies (See Appendix 2 for examples of attention training experiments and exercises).

None of the children received any other form of psychological therapy, though there was an emphasis on all professionals involved in the child's/young person's care (e.g. family/carers, school, allied health professionals) working collaboratively and having a shared understanding of the child's problems and therapeutic intervention being provided. This was facilitated by the Clinical Nurse Specialist working in the service, with liaison provided as needed. Delivery of therapeutic input was individually tailored to meet the child/young person's needs, with consideration of the factors reported in Fig. 1.

2.4. Case vignette (pt 15)

AW was a 15 year old male, with a diagnosis of TS, ASD and OCD. He had a history of chronic fatigue syndrome (from 8 years of age), with separation anxiety as a younger child. He had florid motor and phonic tics, which included explosive speech, coprolalia, facial grimacing, tapping/hitting self and stamping. Some tics were reported to cause physical harm (e.g. falling up the stairs), with him experiencing extended bouts of tics and 'tic-like attacks' (see Robinson & Hedderly 2016 [39]) that contribute to him reporting muscular aches/pains. He had difficulties with impulse control that included rage attacks where items may be broken (e.g. plates, cups), episodes where he would pick up knives and run towards people whilst stating that he will harm them (e.g. brother) and stripping his clothes off and running around naked. AW had been home educated since academic Year 3, due to problems reintegrating at school following a period of illness with chronic fatigue symptoms and frequent absconding.

AW was seen for 10 sessions of psychological therapy for FMD, with consideration of tic related concerns and co-occurring anxiety. A summary of the treatment plan is presented in Table 2.

Following treatment, the tics had settled and were no longer interfering with day to day functioning, though still noticeable when anxious. Functional tic-like attacks had stopped and an improvement in impulse control difficulties was reported. AW

exhibited increased insight into his emotions and a better understanding of anxiety and tic triggers, with him proactively managing these situations by anticipating his attention to what was going on around him and using CBT techniques. Parents reported reduced concerns and increased confidence in him going out independently. He had enrolled in college and had started to engage in a range of social and recreational activities. On the clinician rated CGAS, pre-treatment score = 31 (Serious Problems) and post-treatment score = 78 (Doing All right).

2.5. Data analysis

Since all participants showed reliable change (RCI) a group-level analysis was performed using the Wilcoxon signed ranks test, by comparing total pre- and post-treatment CGAS scores.

3. Results

Eighteen children were seen for treatment (9 female; 9 male), aged between 10 to 18 years, with a mean age of 13 years. The mean age for symptom onset was 11 years (range 6–15 years, $sd = 2.64$), whilst the mean age for presentation to the clinic was 13 years (range 10–18 years, $sd = 2.46$). There was an average symptom duration of 2 years 4 months between age at onset and presentation to the clinic (range 0 years–8 years, $sd = 2.28$). Functional movement symptoms of the cohort included tic-like attacks ($N = 8$), loss of functional movement in legs ($N = 7$), tremors ($N = 3$), fixed flexion ($N = 2$), chronic pain ($N = 1$), visual disturbances ($N = 1$) and non-epileptic attacks ($N = 1$). Co-occurring conditions included TS ($N = 9$), stereotypies ($N = 1$), ASD ($N = 3$), Attention Deficit Hyperactivity Disorder (ADHD; $N = 1$), social anxiety ($N = 6$), obsessive compulsive disorder (OCD; $N = 2$) and low mood ($N = 1$). Non-school attendance was reported for 7 of the cases. The characteristics of all patients and pre-/post-treatment CGAS scores are presented in Table 1.

Patients were seen for an average of 9 individual psychology sessions (range 1–22 sessions). All cases reported clinical improvement in FMD, with 14 cases (out of 18) reporting complete resolution of FMD symptoms. Children for whom symptoms were still reported at follow-up were those with functional tics and one case of a functional tremor; though all reported symptoms only occurring when highly anxious. Three of the cases represented with non-movement related functional symptoms (e.g. headaches, stomach aches, pain). Four cases were identified as presenting with unmet cognitive needs, with neuropsychological assessments conducted to help inform educational provision.

Patients presented with a mean pre-treatment GCAS score of 44.39 ($sd = 8.29$; range = 31 to 62), which was classified as presenting with Obvious Problems. There was a mean post-treatment CGAS score of 73.72 ($sd = 8.89$, range = 60 to 90), which was classified as Doing All Right. All participants showed a significant change in functioning on the basis of the RCI, with clinically significant change across classification boundaries. For the group analysis, the CGAS scores increased significantly from pre-to post-intervention ($p < 0.001$) indicating significantly improved functioning.

4. Discussion

The current article sought to present a cognitive behavioural based approach with attention training components for the management of functional movement symptoms in children and young people. Following an initial multidisciplinary assessment and clear diagnosis of symptoms, eighteen children attended psychological therapy sessions. The children presented with a range of FMDs,

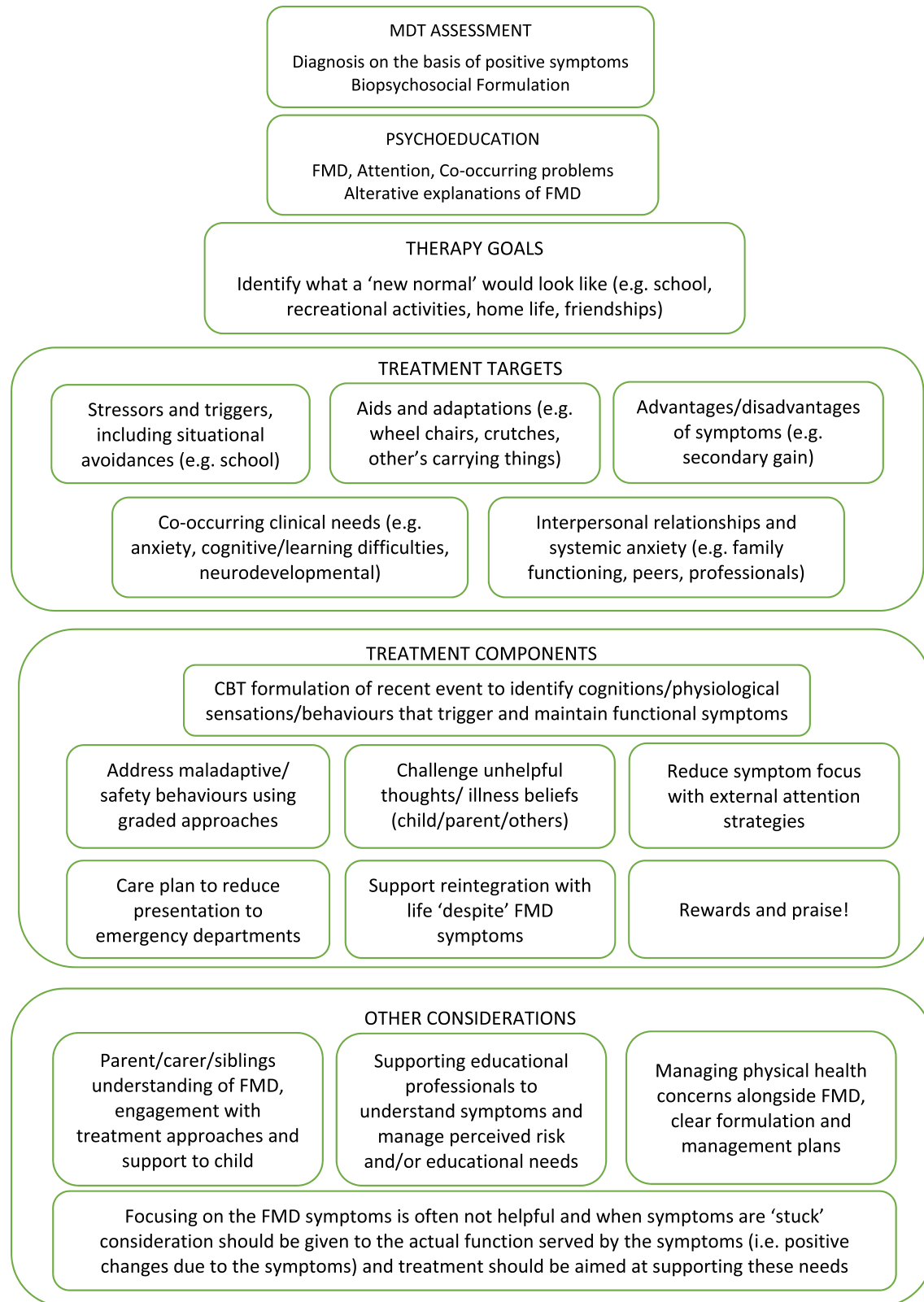


Fig. 1. Treatment components.

including functional loss of walking, tic-like attacks and non-epileptic seizures. A biopsychosocial approach was adopted and consideration was given to the negative and positive impact of functional symptoms, beliefs about the symptoms and co-occurring

mental health and/or educational needs. The treatment was individually tailored to meet the child's needs; however, the same approach was used irrespective of presenting functional symptoms. All children reported an improvement in FMD and clinical

Table 2
AW treatment plan.

Session and Aims	Key Components
Sessions 1–2: Psychoeducation and goal setting	<ul style="list-style-type: none"> • Understanding FMD, tics and anxiety • External attention experiments (e.g. hand experiment) • Advantages/disadvantages of symptoms (e.g. being left alone, sleeping less, missing college, not playing rugby) • Treatment goals (attending college, going to music gigs)
Sessions 3–4: Role of attention (self/others) and formulation	<ul style="list-style-type: none"> • Behavioural analysis/monitoring of tic-like attacks • CBT formulation of tic-like attacks, with consideration of attentional focus, triggering thoughts (e.g. I'm going to have one) and safety behaviours (e.g. parents putting cushion under head, standing at bottom of stairs) • External attention practice (e.g. daily listening to sounds)
Sessions 5–8: Behavioural experiments and thought challenging	<ul style="list-style-type: none"> • Role of attention on symptoms (e.g. performing daily tasks whilst focusing attention on symptoms/away from symptoms such as by humming/singing) • Shifting attentional focus (e.g. walking up/down stairs whilst focusing externally by thinking about the purpose of going up/down stairs) • Graded exposure (e.g. going into town, music gigs, college interview, catching bus)
Sessions 9–10: Review and relapse prevention	<ul style="list-style-type: none"> • Review of goals and treatment gains • Plan for the future

improvement in functioning on the CGAS, with the majority experiencing complete remission of the FMD.

The treatment focused on reducing attentional focus to the symptoms and supporting re-engagement with life, despite the FMD, using psychological approaches. For these reasons we did not include any motor rehabilitation or physiotherapy in the management plan (with only one case receiving local physiotherapy input). The aim of the attention training techniques was to support patients to shift their attention away from internal sensory processes (interoception) and on to the external stimuli (exteroception), with consideration given to the cognitive processing load (i.e. needed to be sufficient to reduce processing of sensory phenomena). There was an emphasis on helping family members to support children/young people to use external attention focusing strategies and to encourage them to be focusing externally when engaging in behavioural experiments. Families reported that this was a comprehensive, achievable and acceptable strategy, with children and parents finding attention shifting techniques effective in reducing the symptom burden and improving participation in life.

The number of sessions delivered varied considerably. For the majority of cases the number of sessions required was in keeping with that proposed for a typical course of CBT (i.e. 8 to 12 sessions). However, for some cases, typically those with a shorter duration of symptoms, psychoeducation about FMD and discussion of strategies to reduce internal attentional focus was sufficient to support symptom management. There were also a couple of cases that required more than 20 sessions; one of these children had the longest symptom duration (8 years) and the other presented with co-occurring chronic pain. These findings are in keeping with previous research that has shown how functional symptoms may become more treatment-resistant with time [6]. It also highlights the importance of good psychoeducation and how including information about the role of attention on functional symptoms may increase understanding of the mechanisms and support families' abilities to engage in more adaptive behaviours.

The impact of symptoms on functioning was significant, with 38% of the sample not attending school when they presented to the service. To support school reengagement, a graded approach was adopted with careful consideration of the reasons for non-attendance and how this may have contributed to the onset and maintenance of functional symptoms. Concerns typically included worries about experiencing FMD symptoms in front of peers, academic difficulties due to unmet learning/cognitive needs (with three requiring additional neuropsychology assessments) and/or school's concerns about the management of FMD and keeping the child safe. The aim was to support children to return to school as soon as possible, even if only for break time, with remedial support

provided to facilitate this (e.g. attending learning support for certain lessons, lift passes/leaving classes early to avoid the rush of students etc.). Liaison with the school (over the phone and/or via email) was often required so that a consistent approach could be adopted, with monitoring of the effectiveness of additional provision and the gradual removal of external aids that might have been implemented to support school reintegration.

Of the eighteen children/young people seen for treatment, six were also seen for psychiatric evaluation of their mood, with three prescribed medication for mood stabilisation. The impact of medication on symptoms is unclear, though the purpose of pharmacological support would be to support engagement with psychological therapy. Despite resolution of functional symptoms in these cases, two presented with persistent mental health needs and were referred to local services for ongoing support. Co-occurring mental health conditions were commonly identified either at clinical assessment or were unmasked as the FMD resolved. This is in keeping with the literature, where co-occurring mental health symptoms have been reported for approximately 25% of cases [3]. It also supports the hypothesis that FMD symptoms may reflect a form of emotional regulation and atypical connectivity between motor, emotional and frontal neural networks [19].

This case series provides support for the use of CBT with attention training components for the management of FMD. Given that all children exhibited a reduction in the FMD and clinically significant improvements in functioning, it can be posited that psychological support and reduced attention to FMD symptoms is a helpful treatment model for children. This is of relevance to health care providers and referrers, as it provides support for the use of psychological therapy for the management of FMD, which should be taken into consideration when developing new services for these complex cases.

4.1. Limitations

The current case series presents a treatment approach developed by clinicians working in a tertiary movement service, with children/young people typically presenting with FMD symptoms in the context of co-occurring neurodevelopmental movements (i.e. tic disorders). The generalisability of the current findings to children/young people with a broader range of functional movement symptoms is therefore unclear (e.g. pain, numbness, paralysis). However, changes in attentional control processes have been proposed to contribute to the onset and maintenance of functional symptoms, so it appears plausible that the current treatment approach would also be acceptable for children/young people with other types of functional movement symptoms. Generalisability

studies that explore the efficacy of CBT with attention training components for children/young people with other functional symptoms would be of interest.

The current treatment was not a manualised approach and the sample included children/young people who were motivated and accepting of the FMD diagnosis, which may have contributed to the positive treatment outcomes. As an open label study, with no comparison control group, the efficacy of the treatment relative to no intervention or CBT alone is unclear. However, given the number of children who presented with functional symptoms for a number of years prior to treatment and the resolution of these symptoms following treatment, we would argue that this treatment package is more effective than no treatment for children with FMD. There is complexity in teasing apart the active treatment components as a broad treatment protocol that incorporated a range of techniques shown to be effective in managing psychiatric symptoms was included. But, we believe that the attentional training components provide additional benefit than CBT alone, so future feasibility studies are required to explore this hypothesis with a direct comparison between CBT vs. CBT with attention training.

Due to the paucity of research in this area, there were no specific clinical assessment tools suitable to assess FMD symptoms, with assessment of treatment efficacy on the basis of self-reported improvement in functional symptoms. Standard measures of mental health were also not included as they have been shown to be insensitive to change in this population [34]. Future clinical treatment studies of CBT with attentional training components would benefit from the inclusion of objective measures of interoception and attentional focus. There are a range of tools that are currently being validated in children without neurodevelopmental or mental health concerns, such as the Interoceptive Accuracy Scale or the Body Perception Questionnaire – Very Short Form [40,41]. Validating these tools in clinical populations would be helpful to explore the role of interoceptive processing in children with FMD, with use in clinical treatment studies providing useful information about potential cognitive mechanisms that contribute to the onset and maintenance of FMD.

5. Conclusions

This case series provides promising preliminary evidence for the use of CBT strategies with additional specific attention training components for the treatment of FMD in children. The children in this cohort did not require additional motor rehabilitation to improve motor functioning. Instead, children and parents reported improvement of motor functioning by reducing attention to the symptoms, with CBT with external attention focusing being an achievable and acceptable treatment approach. Larger trials are necessary to identify which individual treatment components are most effective and to better understand and quantify response to treatment. We aim to develop and manualise this treatment protocol.

Appendix 1

Complex Case Template (TANDeM Service).

This template is intended to aid a clinician's consultation with a child/young person who has been referred to their service with multiple or complex symptoms of uncertain aetiology.

Recommendations:

A. Prior to the Consultation.

If the referral letter indicates clinical complexity (i.e. not exclusively but commonly including: Ehlers Danlos syndromes (EDS), chronic pain, chronic fatigue; Paediatric Acute-Onset Neuropsychiatric Syndrome (PANS), Postural Orthostatic

Tachycardia Syndrome (POTS), conversion disorder; functional neurological movement symptoms):

1. Call GP/paediatrician for a full background of the case (if not already supplied)
2. Send a pre-assessment questionnaire to parent/carer and child – and ask “What are you hoping to achieve from this consultation”?
3. Make a list of all involved services and clinicians (thoroughly check all available clinical records)
4. Allow for a 2-h clinic code/consultation appointment, where possible
5. Arrange to see the child and parent/carer as part of an MDT clinic, with nurse support/psychology/wider clinic team present, where possible

B. During Consultation

1. Establish aims of consultation and referral questions from parent/carer
2. Establish aims and referral questions from child
3. (If possible) Interview child without parent/carer and allow space for each to talk
4. Make a problem/symptom list and avoid focus on diagnostic labels
5. Discuss a solution-based approach to care
6. Ensure full developmental history and milestones has been recorded
7. Educational history - Is there an EHCP in place or an application?
8. Psychiatric history –include screening for developmental conditions (ADHD/ASD) and mood disorders (anxiety, separation anxiety, depression)
9. Social history
10. Family history (make note of any similar symptoms in the wider family)

Record detailed conversations. If any concerns have arisen with other teams, take specific details and ask parent/carer to agree for the team to document in clinical notes; to keep communication clear.

If complaints are threatened stay transparent and provide contacts for Patient Advice Liaison Service (PALS). Document all discussions and specific grievances.

Finally, devise a formulation and discuss this fully with parent/carer and child.

C. Post Consultation.

Ensure detailed and accurate documentation.

Ask parents if they feel their aims have been achieved and document their response.

Discuss in MDT and obtain feedback on meeting and communication.

Involve psychology teams if not already, where necessary.

Involve wider MDT/Occupational Therapy/Physiotherapy, if necessary.

Agree plans and options for future care.

Discuss outcome with GP and wider teams to ensure good communication.

Avoid referring on to a different sub-speciality where possible, without full discussion with GP and agreement that the referral is necessary for care and management.

Appendix 2. Attention focusing experiments

Attention focusing experiments can be used to help support acceptance of the diagnosis and the treatment model. Responses can be used to start discussions about the power of attention on the

body part used in the experiment and how these are 'typical' sensations that are normally present in the body. The notion of interoception and attention to internal sensations can then be initiated, with the discussion of attention as a cognitive process (e.g. selective attention, sustained attention, divided attention). If individuals do not report any sensations during the experiments or non-habituation of sensations following the experiments, then the clinician should explore this and ask the participant what they were thinking of whilst completing the experiment. In our experience, where this is the case, it is because the person is not focusing on the sensations in the hand and thinking about something else.

Hand sensations: Children and parents are instructed to hold out their hand in front of them with their palm facing down. They are then instructed to focus their attention on their hands and keep thinking only about their hand. They are instructed to do this for 1 min. The clinician should also take part in this experiment and monitor whether the child is maintaining attention to their hand, with gentle prompts to refocus their attention, if needed. Following the minute, everyone in the room should be asked what sensations they noticed in their hands. This will typically include responses such as fingers twitched, tingling sensations, heavy feeling. After a few minutes, the clinician should ask the child/parent what sensations they now notice, as a way to demonstrate that when attention is shifted away from the body part then the physiological sensations subside.

For patients with a hand tremor, demonstrating the added effects of attention on the hands on the tremor can be helpful – though this is likely to require videoing so that the patient can observe the difference in attention their self.

Floor on Feet: This is an adaptation of the hand sensations experiment, where children and parents to pay attention to their feet and the sensations that they experience of their feet against the floor (e.g. the floor pushing up on their feet, twitching of toes). This can work well for children who have hand/arm related functional movements and already pay significant attention to their hands.

Shock Ball: Playing a game with a 'shock ball', which is a ball that emits small electrical shocks after random time periods, can be a helpful way to demonstrate the power of attention on sensory processing, habituation and symptom exacerbation. For example, the child can hold the ball and be instructed to focus on receiving the shock, with discussions around whether they experience physical and/or thought related changes the more times they receive the shock. Adaptations can include how tightly or loosely they hold the ball and shifting attentional focus to the shock by engaging in dual activities whilst playing the game (e.g. talking, listening to music). The purpose of this experiment is to stimulate discussion around the role of attention and beliefs on sensory processing. Parental and child/young person consent should be sought prior to playing this game.

External attention training: This was based on the meta-cognitive attention training paradigms, and involved asking children to engage in daily attention training practice (5–10 min), where they would be asked to focus their attention on a 1) a single sound in the environment that was close by, 2) a single sound in the environment that was more distal, 3) switching their attention between these sounds, 4) listening to both sounds together. This was practiced in session with patients and given as a homework task. The purpose of this was to increase patients' awareness of attention processing, with this being a mechanism that can be trained and is within their control.

Sensory Grounding Techniques: These techniques were based on those commonly reported as helpful for patients with non-epileptic attacks and can be used in associated with the other external attention focusing techniques. When developing sensory grounding techniques consideration should be given to ensuring

the strategy is associated with a cognitive processing load that engages attention and does not evoke an emotional response. These typically included:

Auditory: listening to music and decomposing it into separate sounds/instruments.

Visual: describing or counting colours, shapes, textures that can be seen in the environment.

Tactile: touching clothing or surfaces close by and describing the texture.

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