

Case report

A therapy-led, multidisciplinary programme for treatment-resistant functional fixed dystonia

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Accepted 19 June 2020

SUMMARY

Functional neurological disorder (FND) describes various neurological symptoms that are not explained by an organic aetiology. The condition has a poor prognosis. Despite this, there is sparse research that informs clinical interventions for FND, particularly when treating functional fixed dystonia. Our article has outlined an intervention for a patient with a treatment-resistant functional fixed dystonia that was informed by a biopsychosocial model, which aimed to rehabilitate the patient's functional motor symptoms. This led to favourable outcomes including restoring full range of movement in the patient's right foot and improvements in routine outcome measurement scores. The patient also described the programme as life-changing and was able to re-engage in meaningful and purposeful activities.

BACKGROUND

Functional neurological disorder (FND) is the second most common reason for a neurological outpatient visit.¹ It describes a variety of neurological symptoms that are not explained by an organic aetiology.² A cohort study found that patients who experienced functional symptoms had on average an increased rate of disability and distress, and were more likely to claim disability-related benefits compared with those with an organic condition.³ While the precise aetiology of FND is not clear, it is best understood by the biopsychosocial model. In clinical practice, FND diagnosis is formulated on the basis of history taking and positive clinical finding ascertained through physical examination. In most cases it is common to conduct further investigations to rule out organic conditions.⁴

One commonly reported symptom of individuals diagnosed with FND is functional dystonia; it has a poor prognosis with improvement in less than 25% of patients.⁵ This symptom often develops suddenly following a minor injury. It normally presents as immobility and demonstrates marked resistance on passive movement. It can also be accompanied by other functional motor and sensory symptoms as well as localised pain. The most common presentation of functional dystonia in adults is fixed foot plantar flexion and inversion.^{6,7}

Previous research has highlighted the benefit of multidisciplinary interventions and physiotherapy as either a concomitant or stand-alone treatment for functional motor symptoms.^{8–11} There is consensus recommendation¹² and randomised controlled trials^{9,10} that demonstrate the efficacy

of physiotherapy for treating these symptoms. However, within these trials, the sample size of patients who present with functional fixed dystonia is limited. At present, the literature for treating functional dystonia only consists of small-scale cohort studies and case reports and none of them have randomly assigned patients to intervention groups.⁷

Mirrors have been shown to be useful for treating functional motor disorders.¹³ In the context of treating functional fixed dystonia, it can provide visual feedback of an abnormal position of the foot, a position often significantly different to the patient's perception.^{12,13} Mirror box therapy differs in that it uses a mirror to 'trick' the brain into perceiving the affected limb position as similar to the other side. This treatment has also shown positive findings for the overlapping condition complex regional pain syndrome (CRPS).¹⁴ A recent evaluation found 20% of patients presenting with fixed dystonia also fulfilled the diagnostic criteria for CRPS.¹⁵

For the purpose of this article, we outlined an 8-week programme, which was delivered at an outpatient neuropsychiatry unit based at the Bethlem Royal Hospital. The specialist unit offered a treatment that was informed by the biopsychosocial model and included an intensive treatment approach that used group sessions, which were facilitated by an assistant psychologist, rehabilitation assistant, clinical psychologist and specialist physiotherapist.

CASE PRESENTATION

A female patient in her early 40s presented with a 14-month history of right-sided weakness, fixed dystonia in her right foot, dystonia in her right hand and fatigue. She had a history of an eating disorder, anxiety and depression. The patient was admitted to Accident & Emergency (A&E) in December 2015 and initially presented with acute onset of left-sided weakness. These symptoms resolved after discharge. However, in March 2016 the patient started to develop new symptoms on the right side of her body, which included the right foot and the hand assuming a dystonic position. These symptoms deteriorated further over a 6-month period, which resulted in her right foot being in a fixed inverted and plantar flexed position and increased weakness on her right side. As a result, the patient depended on crutches to mobilise. She also experienced pain in her right foot and symptoms of anxiety and depression. During this admission the patient was



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To cite: Hsieh Y, Deshpande S. *BMJ Case Rep* 2020;**13**:e235213. doi:10.1136/bcr-2020-235213

given a provisional diagnosis of FND, which was confirmed by a neuropsychiatrist in June 2016.

INVESTIGATIONS

The patient initially presented to A&E in October 2015 with acute onset of left-sided weakness. During the admission several investigations were carried out including an MRI, CT scan and a routine blood test showing no abnormalities, which ruled out an organic stroke.

She presented again to A&E on March 2016, with the onset of new symptoms on her right side. This included right-sided weakness, while the right foot and hand assuming a dystonic position. Several investigations were carried out, including a repeat MRI of the thoracic spine and brain, nerve conduction tests and a blood test for serum copper. Results were all normal.

The patient was then referred onto a neuropsychiatry outpatient service and was assessed there in July 2016. This included a clinical examination and history taking. During the assessment, the patient demonstrated a positive Hoover's sign and there was a discrepancy between the degree of weakness on objective assessment and when observed mobilising. In addition to this, a genetic test was completed and she was trialled on levodopa, which had no effect. This assessment confirmed a diagnosis of FND.

The patient was then referred to a neurophysiotherapy outpatient clinic, which primarily treated organic neurological conditions. Various neurophysiotherapy interventions were used to treat the right functional fixed dystonia. This included muscle electrical stimulation, botulinum toxin, serial casting, ankle and foot orthosis (AFO) prescription, balance exercises and gait retraining.

None of these interventions benefited the patient and she was referred on to receive specialist treatment at the FiND (Functional integrated Neurological Disorder) programme. This therapy-led programme commenced in April 2017 where a specialist neurophysiotherapy assessment found that the patient's right foot was observed to be fixed in an inverted and plantar flexed position with mild swelling and colour change. On palpation, the right foot was cold and tender to the touch around the lateral aspect of the foot and lower leg, along the peroneal muscles. The right foot showed marked resistance to movement and was unable to be passively corrected. The patient experienced hypoaesthesia on the right side of her body and it was worse on her right foot. There were no signs of allodynia.

The patient demonstrated 4/5 strength on the Oxford muscle test scale in her right lower limb except for the ankle and foot, which was 0/5 as there was no active movement. There was a 'give-way' weakness on the right side on manual muscle testing. The patient had developed muscle wasting in her right calf, which was attributed to her right foot being in a fixed position for a prolonged period.

When mobilising unaided, the patient's right foot maintained its fixed dystonic position, and she could only mobilise short distances unaided due to pain localised around the lateral malleolus. The patient normally mobilised with the aid of two crutches, non-weight-bearing on the right foot and wearing an AFO.

DIFFERENTIAL DIAGNOSIS

At the point of referral to the FiND programme, no other diagnoses were considered. Prior to the referral, the patient completed several medical investigations, clinical examinations and history taking. Results found that the patient's symptoms

were consistent with a diagnosis of FND.² The patient exhibited positive clinical findings, discrepancies between objective assessment and when observed in function. This provided clinical evidence of incompatibility between symptoms and other recognised neurological or medical conditions.

TREATMENT

The patient attended a specialised FND treatment programme that ran for 3 days a week for 8 weeks and was attended by six patients. The multidisciplinary programme included a range of group-based interventions: Cognitive Behavioural Therapy (19 hours), specialist physiotherapy (22 hours), planned physical activities such as modified badminton and football (13 hours), and psychoeducation (11 hours). The programme has successfully treated 61 patients and on average they achieved 'much improved' (2.3 average, range 1–4) when scored on the Clinical Global Impression Scale (CGI).

The psychologically informed work aimed to build on the patient's awareness of how precipitating, predisposing, perpetuating and protective factors influenced her presenting problem.^{12 16} This work enabled the patient to explore her belief systems, how thinking errors maintained behaviours, and how this may have a conditioning effect on functional symptoms. In doing so, the patient had the opportunity to reappraise her thinking errors (ie, magnifying, catastrophising their symptoms) and consider some of the unhelpful behaviours that were maintaining these symptoms. It also showed how exposure to activities (such as physiotherapy) over a prolonged and repeated period exposed them to the role of habituation that can retrain an innate response and gradually introduce adaptive behaviours.

The aim of the physiotherapy sessions was to use distraction techniques while engaging the patient in functional tasks to help retrain normal movement patterns in the dystonic foot. This treatment approach has been outlined in the physiotherapy consensus recommendation, which does not advocate for impairment-based exercises. This approach is typically used when treating organic neurological conditions.¹²

Guided by the consensus recommendation, the patient's AFO was removed prior to her commencing physiotherapy on the education that it would inhibit the restoration of the foot and ankle muscles. The patient was advised to continue using the crutches to mobilise while non-weight-bearing on the right foot until it was able to maintain a neutral position. We also addressed habitual sitting positions with external supports being placed under the medial aspect of the right foot to discourage prolonged periods in end of range.

A graded mirror box therapy programme for the right foot was also implemented during the second week of the programme. The patient was advised to place her right foot into a box, which had a mirror on one side. The mirror was positioned so the reflection of her unaffected foot 'superimposed' onto her affected foot. The patient performed graded exercises with the unaffected foot, starting from just looking at the reflected image to performing a range of movements using a graded approach. The patient took the mirror box home and performed the exercises twice a day for 20 minutes while attending the programme. In addition to this, the patient was provided with a home exercise programme, which consisted of exercises that were performed in the physiotherapy sessions. The patient was encouraged to perform them when not at the programme and these exercises varied from week to week depending on the patient's progress.

The patient engaged in daily relaxation techniques (ie, mindfulness) as one of the coping strategies for her anxieties.

Table 1 Outcome measures

	Assessment	Discharge	2-month follow-up	6-month follow-up
Clinician rated outcomes				
10-metre walk test (m/s)	0.25*	0.45†	1.00†	1.13†
Clinical Global Impression Scale		1.00		
Patient rated outcomes				
Beck Anxiety Inventory	11.00	4.00	6.00	
Beck Depression Inventory – 2	16.00	5.00	18.00	
Work and Social Adjustment Scale	19.00	0.00	7.00	

*Elbow crutch.

†Unaided.

OUTCOME AND FOLLOW-UP

During the 8-week programme a battery of clinician rated outcome measures (CROMS) and patient rated outcome measures (PROMS) (see table 1) were administered at initial assessment and discharge. At the 2-month follow-up session PROMS and the 10-metre walk test were administered and at the 6-month follow-up session, the 10-metre walk test was administered.

The patient demonstrated significant improvements throughout the programme. In particular she achieved a neutral position in her right ankle within the first 3 weeks, which was not possible previously despite her receiving various treatments prior to attending the programme as outlined in investigations. The patient's rapid and significant recovery at this stage is perhaps related to several factors, including an intensive, specialist treatment, which was informed by a biopsychosocial model.

The multidisciplinary team continued to offer the patient treatment over the remaining 5 weeks as they agreed that attending the remaining programme would have a positive impact on maintaining her recovery and prognosis, as well as posing as a positive role model to the other patients on the FiND programme. The patient continued to make improvements, which were reflected in her outcome measures at discharge. On completion of the programme the patient's right foot returned to neutral alignment (see figures 1 and 2) and demonstrated full

**Figure 1** Initial presentation of right foot.**Figure 2** Presentation of right foot at discharge.

range of movement in her ankle and foot. Her strength in her right lower limb improved, scoring 5/5 on the Oxford muscle test except for right ankle plantar flexion, which was 4/5. She no longer experienced pain in her right foot and reported improved sensation on her right side, particularly in her right foot. As a result of these improvements the patient achieved her long-term goal of being able to mobilise unaided indoors and in the community without the support of an AFO and was able to return to driving an unadapted car.

These outcomes also suggest there was a substantial improvement in the patient's quality of life, which was reflected by CROMS and PROMS that were administered at discharge (see table 1). This included the disappearance of mild symptoms of depression and anxiety, and reduced perceived difficulties in undertaking daily activities. The patient increased her participation in voluntary work at the church and no longer perceived difficulties from her symptoms when caring for her family. The CGI Score also indicated her symptoms had very much improved.

During the 2-month follow-up session, the patient displayed further improvements as she demonstrated increased speed during the 10-metre walk test. The positioning, active range of movement and strength of the right foot was maintained 2 months after discharge. Despite this, the patient relapsed and experienced mild symptoms of depression, as well as perceived difficulties in undertaking activities of daily living. This could be due to the adjustment she experienced from no longer attending the programme. Her anxiety symptoms remained minimal at follow-up. This could be a result of the patient using coping strategies she learnt on the programme.

At the 6-month follow-up session, there were further improvements in the patient's speed when performing the 10-metre walk test. The patient continued to mobilise unaided with her right foot maintaining a neutral position. She also continued to drive a car without any adaptations and she was able to do this more frequently and for longer distances.

The patient reported that the programme changed her life and these positive gains were in part a result of the normalising effect of undergoing a treatment with a group of patients with the same condition. The patient established meaningful relationships with the other attendees, which offered motivation for change. After discharge, the patient maintained these friendships, which may have played a role in maintaining her physical and mental well-being.

DISCUSSION

Our case study finds that after initial unsuccessful interventions, our multidisciplinary treatment approach led to favourable outcomes for the patient. This included full restoration of active movement in the right ankle, independence with mobility and improvements in outcome measures. Our results support the limited evidence for the therapeutic benefits of a multidisciplinary approach for treating functional fixed dystonia^{5–7} and the value of drawing from various evidence-based therapies that are informed by a biopsychosocial model.¹²

A possible explanation for these favourable outcomes could be from the use of specialist treatment approaches. The psychological work may have facilitated motivation for change and their willingness to engage in physiotherapy sessions. Our specialist physiotherapy input conceptualised functional motor disorder as an involuntary process that is a learnt movement pattern and driven by abnormal self-directed attention. Physiotherapy sessions therefore used specific distraction techniques to assist the retraining of normal movement patterns.^{11 12} The use of mirror box therapy may have assisted the restoration of function and reduced pain in the fixed dystonic foot through the activation of cortical networks and neural reorganisation through

sensorimotor feedback from the unaffected foot.^{14 17} In addition to this, a group-based setting could have assisted the patient as it may have had a normalising effect of these symptoms and receiving therapy as a group may have provided further motivation for change.^{16 18}

The treatment programme consisted of 65 hours of intensive therapeutic work and the setting of homework. The intensive treatment may have accelerated the process of neural plasticity in response to the therapeutic interventions. This may explain how the patient progressed so quickly within the first 3 weeks. Previous research has shown physiotherapy and psychology interventions can facilitate the reorganising of neural pathways by recruiting new and different neural networks.^{19 20}

Previous interventions for the patient were unsuccessful in treating her motor symptoms, which could be due to them not drawing from a biopsychosocial model, but rather from a passive approach. They did not include psychological approaches in their treatment, thus limiting the patient's exposure to how predisposing, precipitating and perpetuating factors may influence her presenting problem. Exploring these factors plays an important part in recovery and there is sufficient evidence that highlights the link between psychological factors and symptom exacerbation.^{12 16} In our case the patient had symptoms of anxiety and depression, which may have contributed to the symptoms.

In conclusion, our case study showed that after initial unsuccessful interventions, an intensive 8-week outpatient multidisciplinary treatment programme led to favourable outcomes for a patient presenting with treatment-resistant functional fixed dystonia. The patient was able to restore full movement in the right foot and became asymptomatic to pain while mobilising without aids. The patient expressed satisfaction from her increased participation in meaningful and purposeful activities. The patient was grateful for the interventions she received from the FiND programme as they had a positive, life-changing impact.

Acknowledgements The authors thank their colleagues from South London and Maudsley who supported and encouraged the authors in writing this manuscript and also those who delivered the programme. The authors also thank those who read the manuscript and provided their suggestions. In particular the authors thank Dr Johannes Dalhuijsen, Dr Brendon Stubbs, Dr Varsha Deshpande and Shalmali Deshpande.

Contributors YH assessed and treated the patient. She also co-wrote and edited the case study. SD co-wrote and edited the case study.

Funding The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

Competing interests None declared.

Patient consent for publication Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

Patient's perspective

I was very apprehensive starting the FiND programme. Previously I only had physiotherapy combined with Botox injections and a foot splint to try and correct my functional fixed dystonia. This approach did not work and I was therefore referred to the FiND programme at the Bethlem Royal Hospital. I could not weight bear and had been using crutches for 16 months. After a slow start, by the third week I regained some movement in my right foot. I attribute this to the intensive physiotherapy and the different type of exercises provided, including the use of mirror box therapy. These had a positive impact on gaining movement in my right foot.

I strongly believe being treated by a multidisciplinary team, having CBT and psychoeducational sessions helped me understand and cope with functional neurological disorder (FND) better. This has also contributed to my amazing progress in regaining full movement in my right foot. It was very helpful being treated in a small group with other FND sufferers as we were able to support and encourage each other. I left the FiND programme being able to walk unaided. Having the freedom to move about without the use of crutches was a joy and I now can drive again.

Learning points

- ▶ While there are randomised trials that demonstrate the efficacy of treating functional motor disorders, none of these trials assigned patients with functional fixed dystonia to the intervention group. Thus, the empirical evidence for treating functional fixed dystonia is limited.
- ▶ Our case study demonstrates that an intensive 8-week outpatient multidisciplinary treatment programme within a group setting led to favourable outcomes to a patient with a treatment-resistant functional fixed dystonia.
- ▶ Our case study highlights the value of specialist physiotherapy input in a multidisciplinary team for treating functional fixed dystonia.

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