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Case Report

Functional Movement Disorder; Importance of Proper Diagnosis and Treatment: A Case Report

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Abstract

Background: Dystonia is a common movement disorder with a wide range of aetiologies. Delays in the identification and initiation of effective treatments should be minimized to improve patient pain and optimize outcomes. This case report aims to underscore the successful treatment of chronic dystonia with the use of mood-modifying serotonin and norepinephrine reuptake inhibitors (SNRI), and encourage clinicians to consider a diagnosis of functional (psychogenic) movement disorder in patients with dystonia that is refractory to usual treatment.

Case Report: This case report describes a 40-year-old woman who presented to a chronic pain clinic for pain related to cervical dystonia with associated head tremor. Her symptoms were refractory to nearly a decade of quarterly botulinum toxin injections. Based on careful evaluation of the patient's history, a normal neurological examination, increased Generalized Anxiety Disorder Scale (GAD-7), Patient Health Questionnaire-9 (PHQ-9), and Injustice Experiences Questionnaire (IEQ) scores, and unsuccessful symptom management with botulinum toxin A, a diagnosis of functional movement disorder (FMD) was made. Low-dose Cymbalta was initiated. The patient achieved near complete symptom remission and resolution of her chronic pain within 2 months and achieved near complete resolution in 2 years.

Conclusion: A diagnosis of FMD should be considered in all patients with dystonia, but especially in patients who respond inadequately to botulinum toxin injections or other rehabilitation therapies. The treatment of comorbid psychiatric conditions can result in substantial benefits and remission from dystonia due to FMD.

Keywords: Dystonia; Torticollis; Functional Movement Disorder; Dystonic Disorders

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Background

Dystonia is a type of hyperkinetic movement disorder characterized by involuntary, sustained, and patterned muscle contractions that result in abnormal postures (1). Cervical dystonia, known colloquially as torticollis, is the most common focal dystonia (2). This type of dystonia affects the neck and results in either forward flexion, extension, lateral flexion, or cervical spine rotation, depending on the muscle(s) affected (1). Approximately, 75% of patients with cervical dystonia have neck pain, and patients may or may not present with an associated directional head tremor (1). Other dystonia distributions include segmental (affecting one limb), multifocal (affecting more than one limb), hemidystonia (affecting one hemibody), and generalized dystonia (commonly involving the trunk and/or both lower extremities) (3).

Dystonias can be idiopathic, inherited, or acquired, and associated signs or symptoms can be useful in guiding the practitioner toward a specific etiology (1). Dystonias occurring in the absence of any other signs or symptoms are referred to as primary dystonias (3). In contrast, dystonias that present with associated features, such as ataxia, spasticity, or paresis, are termed combined dystonias, and can signify the presence of an organic source (3).

The etiology of dystonia can be difficult to ascertain, resulting in delays in both identification and the initiation of effective treatment (4). A frequently missed etiology of dystonia is psychogenic dystonia or functional movement disorder (FMD). FMD is a subtype of functional neurological symptom disorder (FNSD) (previously known as a subtype of conversion disorder). The manifestations of FMD vary widely, with the most common being functional tremor and dystonia (5). In movement disorder clinics, the prevalence of FMD is estimated to be as high as 20%, yet many cases go unrecognized (5-7). Unfortunately, because delays in the diagnosis and treatment of FMD are common, the prognosis for the remission of FMD is typically poor (5). We consider it important for primary care physicians, physiatrists, neurologists, and chronic pain specialists to be aware of FMD as a cause of dystonia to support more timely diagnoses, reduce unnecessary investigations, and facilitate effective treatment. Importantly, the diagnosis of FMD should be considered in patients with dystonia that do not respond adequately to therapeutic botulinum toxin injections.

Features of FMD: FMD most commonly affects women and tends to occur during adulthood (5). FMD is characterized by unwanted movements, such as spasms, shaking, or jerks involving any part of the face, neck, trunk, or limbs, that are caused by underlying stress or a psychological condition. FMD has various phenotypic forms, with tremor occurring in up to 50% of patients (7). Other FMD presentations ordered from most to least common are dystonia, myoclonus/jerks, gait disorder, tics, and parkinsonism (6, 7). Symptoms often develop abruptly after a precipitating event such as an injury, medical procedure, or emotional event and rapidly progress (5-7).

Table 1. Functional neurological symptom disorder (FNSD)

	Diagnostic criteria
DSM-V criteria for FNSD symptoms	One or more symptoms of altered voluntary motor or sensory function
	Clinical findings provide evidence of incompatibility between the symptom and recognized neurological or medical conditions.
	The symptom or deficit is not better explained by another medical or mental disorder.
	The symptom or deficit causes clinically significant distress or impairment in social, occupational, or other important areas of functioning or warrants medical evaluation.
Types of symptoms	Specify symptom type:
	With weakness or paralysis
	With abnormal movement (e.g., tremor, dystonic movement, myoclonus, gait disorder)
	With swallowing symptoms
	With speech symptoms (e.g., dysphonia, slurred speech)
	With attacks or seizures
	With anesthesia or sensory loss
	With special sensory symptoms (e.g., visual, olfactory, or hearing disturbance)
	With mixed symptoms
Acute or persistent	Specify if:
	Acute episode: Symptoms present for less than 6 months
	Persistent: Symptoms occurring for 6 months or more
Presence of psychological stressor	Specify if:
	With psychological stressor (specify stressor)
	Without psychological stressor

DSM-V: Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition; FNSD: Functional neurological symptom disorder

In fact, precipitating events can be identified in up to 80% of patients with FMD (5). The presence of comorbid pain, fatigue, cognitive symptoms, or neurological disease is common, and symptoms are often aggravated by pain, fatigue, stress, or anxiety (5-7). Patients may describe “good and bad days”, and symptoms may change over time or spontaneously remit (7). Symptoms worsen with attention to symptoms and resolve with distraction (5). Tremor entrainment is a key physical examination finding in functional tremor (7). For example, a head tremor may demonstrate entrainment when the patient is asked to mimic a set frequency of forearm pronation and supination (7).

Signs of functional weakness and non-dermatomal sensory deficits may also be present on physical examination (7). Key features of FMD include the presence of inciting event in 80% of patients, sudden onset of symptoms, symptoms resolution with distraction and enhancement with attention, variability of amplitude, frequency or distribution of symptoms over time, sensory findings not conforming to a physiologically plausible pattern, and signs of functional weakness. The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-V) criteria for the diagnosis of FNSD are outlined in table 1 (5).

We report the case of a 40-year-old woman who was referred for assessment and treatment of chronic pain related to a persistent dystonic neck tremor. For 8 years, this patient had been receiving quarterly Botox injections for symptom management. With repeated injections, the patient experienced transient improvement in her dystonia, with no lasting benefit. Conversely, following our diagnosis of FMD and subsequent treatment with serotonin and norepinephrine reuptake inhibitors (SNRI), this patient experienced complete, long-term symptom resolution. Here, we describe the clinical importance of considering dystonia as a symptom of FMD and highlight the diagnostic benefit of a trial of SNRI medication in further differentiating FMD from organic or primary dystonia.

Case Report

A 40-year-old right-handed woman receptionist was assessed at a chronic pain clinic for neck pain related to dystonia. Her past medical history was significant for transient Bell’s palsy as a young adult, resolved gestational diabetes, anxiety, and previous motor vehicle accidents without sequelae.

The patient described a sudden-onset "head tremor" in early adulthood that was initially intermittent and situation-dependent before becoming constant. Over the course of 10 years, she underwent a computed tomography (CT) scan of the head, a magnetic resonance imaging (MRI) of the head, and a total of three electroencephalograms (EEGs), all of which had no significant findings. She was diagnosed with cervical dystonia and treated for the following 8 years with quarterly therapeutic Botox injections. Immediately following each injection, the patient experienced transient improvements in pain relief and dystonia for 2-3 months at a time, but her cervical dystonia and associated neck tremor never completely resolved.

This patient was subsequently referred to a chronic pain clinic due to chronic neck and shoulder pain from constant head “shaking” and “bending to the side”. Her pain was described as sharp, shooting, and throbbing and was rated as an 8/10 in severity on average. Her dystonia worsened with work stressors and was not present at night. The patient had difficulty staying asleep, feeling unrefreshed in the morning with constant fatigue. Morning headaches occurred frequently. She had no unintentional weight loss nor concerns with bowel or bladder function. The patient described her mood as "fluctuating, mainly emotional and short-tempered”. Her only medication was cyclobenzaprine 10 mg once daily at bedtime. No allergies were reported. The patient was a non-smoker and endorsed occasional alcohol use. She denied the use of recreational drugs. She lived at home with her husband and daughter.

The following questionnaires were completed at the initial visit:

- Pain Stages of Change Questionnaire (PSOCQ) showed the highest score in Part A (precontemplative stage),
- The pain interference score of the Brief Pain Inventory (BPI) was at 22/70 (indicating low levels of pain interference),
- Generalized Anxiety Disorder Scale (GAD-7) was scored at 18/21 (suggesting severe anxiety),
- Patient Health Questionnaire-9 (PHQ-9) was scored at 12/27 (suggesting mild to moderate depression),
- The Injustice Experiences Questionnaire (IEQ) was scored at 20/48 (not indicating a high level of perceived interference).

On physical examination, no pain behaviors were noted. A mild head tremor was evident and constant. Gait

was normal and the patient was able to walk on her toes and heels. Cranial nerve testing was unremarkable, and dermatomes and myotomes were normal. Deep tendon reflexes were 2+ and symmetrical. Cervical spine range of movement was full and pain-free. Spurling's test and Lhermitte's sign were negative bilaterally. The patient reported diffuse tenderness of the posterior neck and shoulders bilaterally on palpation.

No further investigations were performed, and the patient was diagnosed with cervical dystonia due to anxiety (psychogenic movement disorder or FMD).

The following treatment was initiated:

1. Cymbalta 30 mg orally once daily for 2 weeks, increasing to 60 mg for 2 months
2. Botox 200 units were injected for tremor relief (in her upper trapezius, middle trapezius, splenius capitis, sternocleidomastoid, levator scapula, and paracervical muscles bilaterally)
3. Referral to psychologist for stress management techniques and psychological support.

The expected outcome was an improvement in pain and tremor.

Follow-Ups and Outcomes: The first follow-up was two months after the initial visit. The patient's neck pain and headaches completely resolved. According to the patient, her dystonia and head tremor improved by 85%-90%. The patient reported feeling better than she had in 20 years and became "a new person". She successfully returned to work. Her sleep improved and she felt refreshed in the mornings. Her mood was described as "fine" and her anxiety improved significantly. She did not start psychology/counselling. Education was provided on distraction, mindfulness, and pain management, and regular stretching was encouraged.

The second follow-up was one year after the initial visit. The patient continued taking Cymbalta 60 mg once daily, and her pain and tremor remained 90% improved for 1 year. Her tremor returned, coinciding with an increase in work-related stress and her mood was described as "down". The patient had no recurrence of dystonia since first starting Cymbalta. The patient was interested in additional Botox injections. 200 units of Botox were injected for tremor relief (upper trapezius, sternocleidomastoid, scalenes, and levator scapula bilaterally). Outpatient care was discontinued.

Now 2 years since the initiation of Cymbalta, the patient has had no recurrence of dystonia and no reported side effects of Cymbalta. Successful treatment of primary dystonia, newly diagnosed as FMD, was achieved with Cymbalta titrated to effect and 2 rounds of Botox injections occurring one year apart. The summary of follow-ups and outcomes has been provided in table 2.

Table 2. Summary of follow-ups and outcomes

	Botox	Cymbalta (duloxetine)	Psychology	Physical activity
Initial consultation	200 units	30 mg daily for 2 weeks –60 mg daily for 2 months	Recommended	-
Follow-up #1 after 2 months	-	60 mg once daily	Recommended	Encouraged
Follow up #2 after 1 year	200 units	60 mg once daily	-	Encouraged
Outpatient care discontinued				

Discussion

This case highlights several key features that support a diagnosis of FMD rather than dystonia due to an organic or idiopathic cause. First, the abrupt onset of this patient's symptoms is in direct contrast to the gradual onset of symptoms seen in most neurological disorders. Second, an episode of transient Bell's palsy in young adulthood may represent the inciting event that led to the appearance of dystonic symptoms. Third, this patient's completely normal neurological examination suggests a non-organic cause of her symptoms. Fourth, this patient's immediate positive response to Botox injections is not in keeping with the expected response of organic dystonia to botulinum toxin. Botox requires a minimum of 12 to 72 hours to result in neuromuscular blockade (8). Therefore, immediate resolution of dystonia with injections of botulinum toxin cannot be expected of dystonia due to an organic cause. Finally, this patient achieved complete remission of dystonia and associated tremor with low-dose Cymbalta and concomitant improvement in anxiety. This positive response to a mood-modifying SNRI is in keeping with FMD, not organic dystonia.

FMD is treated using a multidisciplinary approach. Patients may benefit from rehabilitation in the form of physiotherapy, occupational therapy, or speech-language pathology, with or without psychotherapy (5). Rehabilitation techniques include distraction, physical activity, mobility aids, and reinforcement of adaptive movement patterns and postures. A psychiatry referral may be helpful to optimize the treatment of comorbid psychiatric conditions like anxiety, depression, post-traumatic stress disorder (PTSD), personality disorders, or psychosis (5). Because comorbid psychiatric conditions can exacerbate FMD symptoms, cognitive-behavioral therapy (CBT) and mood-modifying medications, such as Cymbalta, can be of great benefit. In fact, this case highlights a subjective and objective resolution of chronic dystonia and tremor with regular low-dose Cymbalta, compared to the previous transient and subjective improvement in symptoms with quarterly Botox injections.

Conclusion

FMD is a recognized cause of dystonia, yet the diagnosis of FMD frequently goes unrecognized in patients. The prevalence of FMD in movement disorder clinics is thought to be high, with some estimates as high as 1 in 5 patients. As a clinical diagnosis, the identification of critical features such as an inciting event, abrupt onset of symptoms, symptom variability and distractibility, functional weakness, and other physical examination findings that do not conform to an alternate neurological disorder is helpful in identifying FMD. Improvement in symptoms with an SNRI, such as Cymbalta, can occur in FMD but is unlikely in organic dystonia. Especially in patients with inadequate response to botulinum toxin injections or other rehabilitative therapies, the possibility of a diagnosis of FMD should be considered. The treatment of comorbid psychiatric conditions via psychiatrist referral, psychotherapy, or the initiation of antidepressants or anxiolytic medications may result in a substantial benefit and potential remission of dystonia due to FMD.

Conflict of Interest

The authors declare no conflict of interest in this study.

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