Functional (Psychogenic) Dystonia

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Functional (psychogenic) neurological disorders are common throughout neurological practice, and functional movement disorders (FMDs) form a considerable proportion of these cases. In this chapter, we will consider the entity of functional (psychogenic) dystonia.

8.1 Terminology

A number of different words can be used to describe the broad topic of this chapter. The word used for centuries – hysteria – has now almost completely been abandoned given its connotations that the source of the problem is in the uterus and its lay use as an insulting term [1, 2]. However, one of the most eminent movement disorder specialists of recent times, David Marsden, asserted that neurologists "have clung on to hysteria because its modern roots sprang from neurology" [3]. The term "psychogenic" is the one in most common usage among movement disorder physicians and is one of a number of terms (conversion disorder, somatization, psychosomatic) that suggest a primary (even sole) role for psychological factors in the genesis

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of these movement disorders [4]. However, epidemiological studies fail to find the expected high rates of psychological trauma or distress in these patients [5]. In a survey of members of the movement disorder society reported by Espay et al., only 18 % of specialists used the presence of psychological disorder to make the diagnosis, and, interestingly, psychiatrists often sent patients to neurologists clarifying the diagnosis [6].

In place of these terms and other terms that define the disorder by saying what it is not (nonorganic, medically unexplained), it has recently been suggested that the historical term "functional" is the most appropriate [7]. It is certainly a term which is more acceptable to patients than many of the alternatives [8]. There are arguments to support the use of other terms, but while acknowledging these, we will use the term "functional" in this chapter [9]. In doing so, we seek to define these disorders by positive clinical characteristics as movement disorders that are significantly altered by distraction of attention or nonphysiological maneuvers (including dramatic placebo effects) and which are incongruent with movement disorders known to be caused by neurological disease.

8.2 Epidemiology

Functional neurological symptoms are very common, accounting for 16 % of the newly referred outpatients attending neurology clinics [10], and are the second most common issue for neurological outpatient consultation. The most common functional neurological symptoms are nonepileptic attacks and functional weakness [11]. Functional movement disorders (FMDs) constitute about 15 % of those with functional neurological symptoms. In movement disorder clinics, FMDs account for about 2–5 % of patients, but in tertiary movement disorder clinics, such patients account for 20–30 % of cases [12, 13].

The most common FMDs are tremor and dystonia that together account for about 70 % of patients, followed by myoclonus and gait disturbances. Functional parkinsonism, tics, and chorea are rare [12]. Women are more often affected than men, and the mean age at onset is from 37 to 50 years depending on the study selected [12, 13]. Age at onset for patients with functional dystonia is younger than the average for FMD in general: Schrag et al. found an age at onset ranging from 16.6 to 42.8 [14]. FMDs are also seen in children and elderly patients [15, 16]. Functional dystonia, however, seems to be very rare in those over 60 [16].

FMDs are characterized by the level of disability and impairment in the quality of life equivalent to that reported by patients affected by Parkinson's disease [17]. Consequentially, considerable health and social care costs are associated with this group of patients. Indeed, Carson et al. demonstrated that, in a large cohort of patients with functional neurological disorders (1,144 cases), 50 % of cases had stopped working for health reasons and 27 % were receiving disability-related financial benefits. We can estimate that the economic impact is similar for patients with FMD [18].

8.3 Historical Development of the Concept of "Psychogenic" Dystonia

One of the oldest descriptions of cases of "psychogenic" dystonia comes from Allen in his *Observations on the Motor Phenomena of Hysteria*. Historical descriptions of cases of proposed psychogenic dystonia (until at least the 1980s) are complicated by the fact that many forms of dystonia that are now recognized to be organic were classified as psychogenic. However, cases III and VI by Allen are emblematic descriptions, respectively, of functional foot dystonia and hand dystonia. Clinically, he describes these conditions as "tonic paralysis" and "tonic paralysis with contracture" highlighting the fixity of the postures in these cases that over the time can cause permanent contractures [19].

Fahn and Williams proposed diagnostic criteria for psychogenic dystonia and analyzed the clinical characteristics of 21 patients categorized as having either documented ($n=17$) or clinically established ($n=4$) psychogenic dystonia. Seven of these individuals had paroxysmal dystonia. All these groups of patients presented inconsistent or incongruous dystonic movements. Other important clues commonly exhibited were give-way weakness, the complaint of pain and tenderness to touch, "false" sensory findings, multiple somatizations, and inconsistent slowness of movements. The patients with psychogenic paroxysmal dystonia also had nonepileptic attacks (defined so as patients had abnormal movement associated with apparent loss of consciousness). They highlighted the presence of dystonia at rest in their patients, contrasting with the triggering/worsening of organic dystonia by action. The most common site of onset was in the foot, and they highlighted the role of injury and immobilization (casting) as triggers to onset. In several patients, there was a spread of involvement to other parts of the body. The importance of this paper can be judged by the fact that the diagnostic criteria proposed by them were then adopted as criteria for psychogenic movement disorders in general [20].

Sa et al. described 16 patients with "post-traumatic torticollis," highlighting the issue of peripheral trauma in triggering the onset of dystonia in these patients. These patients had fixed abnormal postures of the neck and lacked sensory tricks or response to botulinum toxin injections [21]. The role of peripheral trauma had previously been highlighted in other papers, and then as of now, it was an issue of some controversy with some suggesting that peripheral trauma could cause dystonia via an "organic" route. Authors also highlighted the issue of severe pain, often identical to that seen in "reflex sympathetic dystrophy," now termed complex regional pain syndrome type I (CRPSI) as an accompanying feature. Several case series reported dystonic posture following peripheral nerve injury [22–24] or surgery [13, 25, 26]. Marsden and Schott described cases of fixed dystonia with CRPS following peripheral trauma, and Bhatia et al. described a series of such cases, calling them "causalgia-dystonia." These patients had vasomotor, sudomotor, and trophic changes and typically had a poor response to botulinum toxin and were very disabled. In line with other FMDs, young women predominated in this group [27].

Van Hilten et al. labeled this condition "tonic dystonia of chronic regional pain," highlighting the presence of severe pain associated to fixed postures similar to that reported by patients affected by complex regional pain syndrome type I (CRPSI) and highlighted an organic explanation for such symptoms. In this series, 26 patients with CRPSI progressed toward a multifocal or generalized tonic dystonia. The abnormal posture began distally, involved mainly flexor muscles, and was associated with sensory (hyperesthesia, hyperalgesia) and autonomic symptoms (sudomotor activity, swelling, discolored skin, decreased skin temperature). They reported an association with the HLA-DR13 haplotype in these patients, proposing it to be a susceptibility factor for this distinct phenotype of CRPS associated with tonic dystonia [28].

In the same period, Verdugo et al. argued in favor of a functional explanation of post-traumatic dystonia. In this series, no structural central or peripheral nervous system anomaly was present in any CRPSI patient displaying abnormal postures. Furthermore, they found that every patient presented at least one additional functional motor sign [29].

The largest clinical series of patients is that reported by Schrag et al. They described clinical features of fixed dystonia in 103 patients (84 % female) [13].The fixed postures involved predominantly the limbs (90 %) and rarely the neck (6 %) or jaw (4 %), and in 61 %, the spread occurred to other body parts. Schrag et al. confirmed the high frequency of the temporal association between fixed postures and physical trauma. Indeed, 64 % presented with dystonia after a minor peripheral injury. Many had associated pain and 20 % of patients fulfilled the criteria for CRPSI. In this series, the triggering injury reported was mainly injuries of soft tissues, but limb overuse, fractures, and consequent immobilization in a plaster cast were also reported as precipitating events. In line with previous studies, dystonia developed subacutely over days or weeks and showed typical features of functional dystonia, as emphasized by Verdugo et al. [29].

8.4 Clinical Features of Functional Dystonia

In recent times, there has been a shift in diagnostic criteria for functional neurological disorders in general with an emphasis placed on positive features in the history and examination and a reduction in emphasis on concomitant psychological distress, which itself is present in a great many patients with neurological disease. This process is reflected in newer diagnostic criteria for psychogenic movement disorders, in attempts at formalizing electrophysiological test batteries, and in the latest criteria for conversion disorder reported in DSM-5 [30–33].

The mainstay of positive diagnosis in patients with functional motor symptoms such as weakness and tremor is the demonstration that normal movement can occur when attention is distracted. Thus, Hoover's sign of functional weakness [34] and various techniques used to assess functional tremor (tapping tasks, ballistic movement tasks) [35] all rely on accessing movement in an unattended fashion and demonstrating a significant change in function compared to when the patient's attention is not distracted.

Table 8.1 Diagnostic criteria for FMD [20, 30]

Fahn-Williams criteria	
Category	Criteria
Documented	Persistent relief by psychotherapy, suggestion of placebo response, which may be helped by physiotherapy, or absence of abnormal postures when patient is unobserved
Clinically established	The posture is inconsistent over time, or it is incongruent with classical dystonia (e.g., impossibility to move limbs on request and great resistance during passive movements), plus one of the following manifestations: other psychogenic signs, multiple somatizations, or an obvious psychiatric disorder
Probable	The dystonia is incongruent or inconsistent with typical organic dystonia, or there are psychogenic signs or multiple somatizations
Possible	Evidence of an emotional disturbance
Gupta-Lang criteria	
Clinically definite	Documented category
	Clinically established category
	Incongruent and/or inconsistent movement disorders (even without the additional presence of psychogenic signs, multiple somatizations, or psychiatric disturbance)
Laboratory-supported definite	Electrophysiological tests proving an FMD (primary evidence of premovement potentials before jerks or data from tremor studies)

The discussion of the historical development of the concept of functional dystonia has revealed that the most common manifestation is with fixed abnormal postures. These postures are often not changed a great deal by distraction. This could of course be because they are not an FMD as some have argued, but an alternative explanation is that maintenance of such fixed postures requires very little attentional focus as opposed to functional tremor or weakness. The key diagnostic points are therefore historical features, pattern recognition, and the search for additional functional signs and symptoms (Table 8.1).

Patients with functional fixed dystonia typically present acutely or subacutely after painful injury to the limb/body part where the abnormal posture arises. The posture may emerge after a period of casting of the limb. Postures tend to be fixed and cannot be moved even passively from their position. There is often a paradoxical relaxation of muscles at rest (even though the posture is maintained) and a very strong contraction of muscles as soon as an attempt is made to move the limb. This is often despite the fact that voluntary contraction of the same muscles to command is weak or nonexistent. The distribution of the dystonia is unusual for the age at onset with multifocal/generalized dystonia common despite onset in adult life. There is typically an absence of sensory gestes or task specificity. The typical pattern of hand functional dystonia results in flexion of the fingers at the metacarpophalangeal or interphalangeal joints. Often the last three fingers are more affected than the second one, with the thumb least affected or not affected. Involvement of the lower extremities results in plantar flexion and inversion of the foot and

sometimes additional clawing, curling, or scissoring of toes. Consequent to prolonged maintenance of tonic postures, patients may have muscle atrophy and joint contractures [13, 36].

Fixed dystonia is often associated with other functional movement disorders or other functional neurological symptoms, including tremor and nonepileptic attacks. With distraction, one can sometimes appreciate a sudden giving way of the abnormal fixed posture. There is an absence of "hard" neurological signs such as pathologically brisk reflexes and extensor plantar responses.

Schrag et al. reported higher rates of affective disorder, dissociative symptoms, and somatization disorder in fixed dystonia than in organic dystonia patients [13]. An association between fixed dystonia with or without clinical features of CRPS and joint hypermobility syndrome has recently been proposed [37]. Joint hypermobility syndrome is characterized by joint laxity which causes articular dislocations, subluxations, and arthralgia [38]. In these patients, the clinical pattern often involves urinary dysfunction and postural orthostatic tachycardia syndrome [39].

There are other patterns of functional dystonia, in particular fixed postures affecting the face ("the smirk") [40] and paroxysmal attacks of dystonic posturing. These latter attacks are phenomenologically different from the known patterns of paroxysmal dystonia occurring as part of paroxysmal kinesigenic dyskinesia, paroxysmal non-kinesigenic dyskinesia, and paroxysmal exercise-induced dystonia. They often have multiple triggers, have highly variable attack duration, and are complicated by other symptoms not seen in organic paroxysmal attacks including very severe pain and apparent alteration of conscious level.

8.5 Pathophysiology and Neurobiology of Fixed Dystonia

Historically, concepts of dissociation, conversion, and somatization formed the basis of the diagnosis and pathophysiological explanation of functional neurological symptoms including functional dystonia. Such concepts, while accepting the reality and involuntary nature of symptoms, suggest a key role for psychological trauma in the genesis of such symptoms. In turn, this suggests that the mainstay of treatment should be uncovering and treatment of this psychological trauma.

As mentioned above, the evidence in favor of psychological trauma playing a key role in the pathophysiology of functional neurological symptoms, including functional dystonia, is weak. In addition, purely psychologically based explanations for symptoms leave a large "explanatory gap": how does one go from a psychological trauma to physical symptoms which are experienced as involuntary, and how can this process be implemented within the brain?

A more neurobiologically focused model of the genesis of functional neurological symptoms in general has highlighted three key processes: abnormal self-directed attention (self-monitoring), abnormal beliefs/expectations, and abnormal sense of agency [41]. The first key feature that distinguishes clearly patients with FMD from those with "organic" movements is the abnormal self-directed attention, an essential factor in manifesting symptoms. When attention is distracted,

there is typically improvement of functional movements. Conversely, symptoms get worse during physical examination, during which attention is drawn toward the body. An experimental exploration of the key role of attention toward FMD was conducted by Parees et al. [42], where patients with FMD showed the impaired performance in movements that were highly predicted (a condition in which there is opportunity for attention toward movement production) compared to healthy subjects.

The nature of attention in fixed dystonia has been added recently by a study of regional cerebral blood flow by positron emission tomography [43]. They observed prefrontal activation in functional dystonia as well as in organic forms; it could represent abnormal movement-related attention. An activation of the basal ganglia and cerebellum has been seen only in functional patients, and it could explain additional problems with self-directed attention/self-monitoring or could reflect a contribution from connected limbic structures.

The second key concept in the neurobiological model of FMD is the presence of abnormal (high-level) beliefs related to symptoms. This does not just mean consciously reportable beliefs about symptoms but involves abnormal predictions and expectations in the setting of active inference in the brain, where the brain is proposed to actively predict and seek to explain sensory input on the basis of past experience. Expectations or prior beliefs play an important role in altering sensory experience. Voon et al., through a number of functional imaging studies in FMD, proposed a model of generation of functional symptoms. Emotional arousal events might be a trigger for the development of movements controlled by the supplementary motor area, functionally disconnected from top-down control by the prefrontal cortex [44].

The third pathophysiological aspect is the abnormal sense of agency exhibited by this group of patients. They have movements that have characteristics one would associate with voluntary movement, for example, the requirement for attention for movement abnormalities to manifest, but yet the movements are not associated with a sense that they are under self-control. There is evidence that patients with FMD and motor conversion disorders experience a lack of sense of agency for movement in general, supporting their own self-report that they cannot voluntarily control the abnormal movements they produce [45].

In the realm of functional dystonia itself, several studies have been conducted with the aim of comparing organic and functional dystonia. Starting from the knowledge that organic dystonia is characterized by abnormal cortical and spinal inhibition [46], Espay et al. examined in the affected limb cortical and spinal inhibitory circuits and cortical activity associated with voluntary movements in functional and organic dystonia patients. Surprisingly, they found that these patients had a similar reduction of short- and long-interval intracortical inhibition (SICI and LICI), as well as the same increase of cortical silent period [47]. Consistent with these findings, Avanzino et al., testing both affected and unaffected sides, observed that SICI and silent period were reduced bilaterally to a similar extent in patients with primary dystonia and those with fixed dystonia [48]. While these findings could be explained by there being a similar endophenotypic trait to reduced cortical

inhibition in patients with organic and functional dystonia, an alternative explanation is that there are confounding factors that influenced the results in those with functional dystonia. Reduced SICI and silent period are not specific markers of dystonia. They are abnormal in a number of neurological disorders but are also influenced in healthy subjects by attention toward the limb, muscle contraction itself, and psychological state.

Quartarone et al. found that organic dystonia patients only exhibited an abnormal (excessive) response to a motor cortical plasticity protocol, in comparison to a normal response in patients with functional dystonia [49, 50].

In a recent study, Katschnig and associates explored the pathophysiology of fixed dystonia applying two experimental techniques: mental rotation of body parts and somatosensory temporal discrimination. They observed that patients with fixed dystonia were slower at performing a mental rotation of a body part displayed in different orientations on a screen than healthy controls, showing a similar level of impairment to patients with organic dystonia. Furthermore, only patients with organic dystonia had abnormalities of temporal discrimination [51]. However, a recent study has found abnormal temporal discrimination in functional dystonia. There are, however, clearly possible influences from abnormal attentional focus in such paradigms which are based on self-report [52].

The impairment in the mental rotation task supports the concept that patients with fixed dystonia may have a distortion of body image, since the ability to perform mental rotation of corporal objects is linked to the concept of "body schema" [53]. It has been proposed that aberrant proprioceptive input due to the prolonged abnormal limb posture could produce secondary changes in central body image. Body image disturbance might contribute to pain and other features, in particular driving some patients to seek the amputation of the limb affected, as experienced in our clinical practice [54]. This phenomenon is also seen in patients with "apotemnophilia" or "body integrity identity disorder" [55]. This fits with previous studies regarding amputation sought by patients with CRPSI and is evidence that at least a proportion of these patients reported fixed dystonia [56, 57]. Therefore, fixed dystonia with or without CRPSI could be considered as a type of body integrity identity disorder, in which a peripheral painful stimulus might be the trigger. Further support for the notion of preexisting disturbed central body schema is the almost uniform lack of success of such amputations with the development of chronic pain and functional movement disorder in the stump and spread of the movement disorder to other limbs [58, 59].

Another important area of interest is the pathophysiological link between peripheral trauma and dystonia. Peripheral mechanisms, such as sensitization of peripheral nociceptors or ectopic or ephaptic transmission of nerve impulses, have been suggested by Jankovic et al. [60]. The spread of symptoms in different body parts, even those not involved in physical injury, seen in fixed dystonia cannot be explained by peripheral mechanisms. Impairment of neuronal circuits at the spinal and brainstem level and central synaptic reorganization have been suggested by van Hilten [28, 61].

8.6 Prognosis and Approach to Treatment of Fixed Dystonia

Evidence regarding the most useful approach to treatment is still controversial, but early intervention seems to be the best way in preventing the development of chronic symptoms and irreversible contractures [13]. Delay in management tends to lead to an unfavorable long-term outcome. The first important step in a successful treatment approach is the optimal communication of the diagnosis with rational explanation of physical symptoms, highlighting the positive way in which the clinician reached the diagnosis.

As reported by Schrag et al. [13], pharmacological and surgical treatments are often unsuccessful. Therefore, an important part of treatment is reduction and/or withdrawal of unnecessary medications and avoidance of unnecessary invasive procedures. Antidepressants and anxiolytics may be helpful in those with anxiety and depression [62]. Two studies have investigated the efficacy of intrathecal baclofen in patients with fixed dystonia associated with CRPSI. Initially, a placebo-controlled study done by van Hilten et al showed impressive response to this drug with marked improvement of the postures, especially of the upper limbs, and of the pain and sensory symptoms [63]. The limitation of this study was the small number of patients and the systematic unblinding of subjects that is likely to have occurred given the prominent systemic effects of intrathecal baclofen. A follow-up study of a larger group of patients found beneficial effects in many patients but a high rate of long-term complications [64].

Cognitive behavioral therapy (CBT) may lead to partial or complete remission of functional symptoms [65, 66]. CBT is directed toward how thoughts, beliefs, and expectations influence the occurrence of unexplained symptoms and to identify behaviors that may be contributing to maintenance of symptoms.

Several studies provide evidence that physical rehabilitation is useful to treat functional motor symptoms [67–69]. Such an approach can be combined usefully with non-pharmacological approaches to pain management, including desensitization, graded mobilization, and the use of specialist pain management CBT techniques.

One approach to the management of severely affected patients is inpatient multidisciplinary rehabilitation [13], providing both physical and psychological treatments in a more intensive fashion than can be achieved as an outpatient.

In addition, placebo interventions can have dramatic effects in patients with functional dystonia, but the long-term benefit of such approaches is not known. In this regard, dramatic immediate responses have been reported to botulinum toxin injections [70, 71] and transcranial magnetic stimulation [72].

Prognosis from published studies is generally poor [73, 74]. One long-term follow-up study has reported improvement in less than 25 % of patients, remission in only 6 %, and progressive worsening in one third. In this study, patients with CRPS and fixed dystonia had poorer outcome [75].

Conclusion

Functional dystonia can be an extremely disabling condition. Future studies are required to develop more objective diagnostic criteria to distinguish functional dystonia from other forms. However, with appropriate management, improvement and even resolution of symptoms can occur. There is a clear need for studies that can delineate the most effective treatment strategies and to build these into clear management pathways for these patients.

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