Psychogenic Dystonia

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Abstract—Psychogenic movement disorders pose a complex problem in modern neurology, which requires an interdisciplinary approach to solve a number of questions related to classification, diagnosis, treatment, and rehabilitation. The most frequent forms of psychogenic movement disorders include tremor, dystonia, myoclonus, and gait abnormality. A clinical case of a 46-year-old male patient with a psychogenic movement disorder presenting as fixed hand dystonia not accompanied by pain is reported. The terminology issues related to the most accurate determination of this type of hyperkinesis, as well as clinical tests (standard motor-skill tasks, ballpoint pen writing) that allow one to identify the psychogenic dystonia is thoroughly analyzed, and the differential diagnostic criteria of psychogenic and primary (idiopathic) dystonia are presented.

Keywords: psychogenic movement disorders, conversion disorder, psychogenic dystonia, idiopathic dystonia, differential diagnosis

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INTRODUCTION

Psychogenic movement disorders (PMDs) can present as any known organic movement disorder [1– 3]. Usually, these disorders are manifested as tremor, dystonia, myoclonus, and gait disturbances [4, 5]. Psychogenic dystonia is one of the most difficult-todiagnose conditions in this group [6]. Among patients seeking medical advice for dystonia, psychogenic dystonia occurs in 2.1–2.9% of cases [5].

Pathology of the extrapyramidal nervous system is a very complex field of clinical neurology, which is associated with a number of factors: a variety of manifestations, an individual variability and the dynamism of extrapyramidal disorders, the lack of clear clinical and paraclinical criteria for identification of syndromes and understanding of their nosology, insufficient knowledge of the etiology and pathogenesis issues, and the absence of adequate treatment [7]. Genetic heterogeneity of idiopathic dystonia also contributes to problems in the disease diagnosis [8-11]. Clinical manifestations of idiopathic dystonia include prolonged muscle contractions resulting in persistent repeated twisting movements and pathological postures in the involved area [12]. Dystonia has certain specific features that can be assessed only by clinical examination; routine neurophysiological tests and neuroimaging techniques (MRI) can not be used either for diagnosis or for classification of dystonia. Primary (idiopathic) dystonia, predominantly in the form of dystonic hyperkinesis, accounts for almost 90% of dystonia cases.

The diagnosis of PMD is based on clinical evaluation of a motor phenomenon, detailed analysis of the disease history, and a number of classical signs identified by neurologic examination [13]. The distinctive PMD features include: sudden onset of manifestations, incongruous symptoms, distractibility and suggestibility of the patient, entrainment of new symptoms, lack of response to otherwise effective pharmacological agents (Table 1).

In the literature, there is no consensus on the terminology of these disorders [14]. Some authors prefer to use the terms "functional" or "conversion (hysterical)", others prefer the terms "psychogenic" [1, 15, 16] or "somatoform" [17, 18]. In practice, the term functional/psychogenic movement disorders is used most often [17-21]. Psychogenic movement disorders are coded in the section F45.8 "Other somatoform disorders" of the ICD-10 classification and the updated ICD-10 version (2016) [22] and in the section 300.81 "Somatoform disorders, not otherwise specified" of the DSM-V [23]. According to the currently discussed ICD-11 version [24], psychogenic movement disorders are included in the section 7B4Z "Bodily distress disorders, unspecified". Therefore, the PMD terminology still remains disputable [13, 25].

Psychogenic dystonia	Idiopathic dystonia	
Onset with rest dystonia	Onset with action dystonia	
Adult onset with leg dystonia	Rare involvement of the leg in adult onset dystonia	
Often fixed spasm	Mobile spasm	
Inconsistent dystonic movements over time	Stable over time	
No corrective gestures	Corrective gestures	
Frequent pains intensified by touching and passive motion	Usually occur without pain (exception—cervical dystonia)	
No improvement after sleep	Often, improvement after sleep	
Other psychogenic movement disorders may be present	Other movement disorders may be present	
Other psychogenic neurological symptoms may be present	Dystonia may overlap with other organic movement disorders	
Usually sudden onset	Sudden onset is untypical	
Spontaneous or placebo-induced remissions	Spontaneous remissions in cervical dystonia	
No family history	Often, family history	

Table 1.	Differential diagnostic	criteria for psycho	genic and idiopathic of	lystonia (according to A	A.E. Lang. 1995; abridged) [6]
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In this article, we present a clinical case of a patient with a psychogenic movement disorder presenting as right hand dystonia.

CLINICAL CASE

A 46-year-old male patient A. presented with slow clenching of the right hand for 30-40 s: attempts to hold small dense objects, e.g. a glue stick, were accompanied by stronger cramping of the hand; the patient was not able to hold a plastic cup because he immediately crushed it due to involuntary cramping of the fingers. The patient described the manifestations of his movement disorder as follows: "At night, during sleep, the hand is relaxed, and the fingers are not cramped; but 15-30 min after awakening, all the symptoms develop-the hand cramps". The hand extended painlessly, but with effort. The patient performed all household (eating, washing, etc.), professional (printing), and other actions with his left hand. If he needed to demonstrate something, he unclenched the right fingers with his left hand, and then put an object in them. The patient did not see a relationship between the movement disorder and trauma or stress.

Disease history. The described symptoms developed 5 years ago; before this time, the patient had noted hand tension when writing for 2 years. During the disease, the patient used leather gloves of varying density and thickness as well as a wooden device invented by himself (finger expander) for compensatory purposes and hand resting. According to the patient, painful sensations arose from that "fingernails digging into the palm, so he held his hand either in the wooden device or wore a leather glove while working.

Medical history. At the age of 18 years, during military service in Afghanistan, he had a very severe concussion, a burn of the right arm, right side of the trunk, and skin of the face and head, and a lacerated wound of the right cheek; he was unconscious for about 1.5-2 h. After treating at a hospital, he was discharged, entered an institute, and graduated from it with honors.

Primary examination (February 18, 2015). There were consequences of a closed lacerated wound on the face. The right nasolabial fold was smoothed (consequences of a wide long scar along the nose, from the upper lip to the eye corner); the chin was slightly turned to the right. The patient was emotionally labile and friendly in conversation and expressed interest in the interlocutor.

Neurological status. The patient satisfactorily performed all mimic tests. The sternocleidomastoid muscle at rest was visually insignificantly contracted on the right, at the area of its clavicular origin; palpation did not reveal changes in the muscle tone on both sides; neck movements were not restricted. The Marinescu-Radovici reflex was positive on the right. The range of active arm motion was completely preserved and painless; no atrophy of the limb muscles was revealed. When speaking, the patient helped himself gesturing with his left arm; the right (affected) arm "did not participate" in the general motor skills; in this case, raising of the right shoulder was noted. Muscle tone in the upper and lower limbs was not changed. The muscle strength in the hand was 5 points on the MRC scale; the finger function test in the right little finger revealed slight weakness (4.5 points). It was noteworthy that the crossing of the right fingures 3 and 4 helped the patient to hold the hand in a relaxed and extended state. Tendon reflexes of the arm and legs were brisk; there were no pathological hand and foot signs. The patient maintained balance in the Romberg test and confidently performed coordination tests; there was no hand tremor. Walking was not significantly changed. There were no pelvic organ disfunction.

A video of the patient with idiopathic or psychogenic dystonia is an important stage in the diagnosis; the video may be analyzed again to clarify details of a motor phenomenon [12]. During video recording, we conducted a series of special tests for in-depth clinical and neurological analysis of the patient's movement disorders.

Test 1. Evaluation of a Motor Phenomenon in the Hand when Performing a Motor Task (Test Duration-45 s)

During holding the right hand (at rest) on the table, only the distal phalanx of the index finger was bent separately, and after a few seconds, the distal phalanges of all fingers were bent. According to the patient, "fingers bent by themselves". Bending of the fingers occurred due to isolated flexion of the distal phalanges. When bending the fingers, the patient lifted the hand over the table, spreading his fingers and demonstrating the phenomenon of "sleeping fingers" due to residual tension in the hand. Then, he tried to hold small objects (glue-stick) using a corrective gesture (grip of the right hand wrist with his left hand) reducing flexion of the fingers. After that, he informed a doctor that there was thenar tension on the right, demonstrating that the hand occurred in a "twisted" position like a "clawed paw". In this case, the patient was able to independently close and open the hand.

Test 2. Holding a Soft Plastic Cup (Test Duration—1 min 30 s)

When transferring a cup, the right hand became a "clawed paw". For 10-15 s, the patient fingered the held cup, flexing and then extending his fingers. After that, he periodically moved the right elbow aside. holding his forearm evenly. On the 30th second of the test, the patient involuntarily compressed the cup and held it compressed for about 5 s. In a few seconds, the patient relaxed his fingers and freely held the cup. Then, he again demonstrated enhanced "involuntary" compression of the cup. During the whole test, the patient demonstrated either relaxation of the hand or its excessive clenching. During the test, the patient also demonstrated the effect of a corrective gesture in the form of touching the right hand wrist with the left hand. When holding the cup with the left hand, there was no tone change; in this case, the patient extended the right hand and shaked it freely. In addition, while holding the cup in the hand being in a supination state (palm facing up), there was neither bending of the fingers nor enhanced compression of the cup.

Test 3. Ballpoint Pen Writing (Test Duration—1 min)

At the moment of initiation of writing, the patient immediately held his hand in an unnatural position, strongly bending the thumb and index finger, and began using a corrective gesture in the form of touching/pushing the writing hand or enhanced pressing of the paper sheet with his left hand. Fifteen seconds after the onset of writing, the patient experienced a need to take a break and shake the hand. Immediately after the patient put his hand on the table, a "clawed paw" formed within a few seconds. The patient pressed the right hand to the table and, when extending it, held it as if under a press. In this case, the patient could lift the hand above the table and held it in a bent position, and then again continued writing. The patient reported that he was not able to write for more than 2 min, "the hand got tired and needed extending and shaking". The patient's handwriting was smooth and legible, and letter loops were well formed. Drawing of figures and numbers was not affected: the patient could freely (without tension in the hand) draw small figures (circle, square, triangle) immediately after writing a text.

Additional Tests

Laboratory tests (complete blood count, general urine analysis, and standard blood chemistry) revealed no abnormalities.

Electroneuromyography findings: the nerve conduction velocity in the motor and sensory fibers of the radial, ulnar, and median nerves of the right upper limb was not disrupted.

MRI of the brain and spinal cord did not reveal any pathology.

The patient evaded additional consultations of specialists (including a psychiatrist) without explaining the reasons. He also refused medication.

Re-examination (12.01.2016): complaints were the same; no worsening of the neurological status was revealed. The patient continued working, holding a senior position at the institution.

DISCUSSION

Psychogenic movement disorders result from mental, rather than neurological, disorders [15]. The category of an unspecified somatoform disorder, as in the presented case, is used in cases where the patient presents with long-lasting, unstable, and multiple symptoms, but the complete clinical picture of somatoform disorder is absent [23]. First of all, the clinical picture of PMD in the patient has a number of peculiarities, which additionally complicates recognizing and interpreting the manifestations of this disease [26]. Psychogenic dystonia implies the presence of psychogenic circumstances and characteristic personality disorders in the patient, while hysterical syndromes develop actually in 20% of patients with a hysterical personality [18]; in some cases, applied psychological tests reveal no changes in these patients [27]. Usually, hysterical movement disorders are accompanied by a decrease in muscle tone, lack of reflex asymmetry, and the absence of muscle atrophy. One of the most frequent hysterical neurological phenomena is paresis. In fact, this specific pseudo-paresis may be considered as "local akinesis" or paralysis of motion, rather than paralysis of muscle or muscle groups [18].

The international literature uses the term "fixed dystonia" [28] that includes manifestations of complex regional pain syndrome (CRPS) and psychogenic dystonia [29]. Often, "fixed painful hand dystonia" is associated with trauma [30] or damage to the "affected area" [31], which is usually related to the development of CRPS [28]. Fixed dystonia of the right hand more often occurs in females, ranging from 51% [32] to 85% of cases [29], with more than 50% of these patients having a certain neuropsychic disorder. Most often, an abnormal hand posture is represented by flexion of the fingers at the interphalangeal or metacarpophalangeal joints. Usually, fingures 4 and 5 are most involved in the fixation, while the thumb is almost never involved in hyperkinesis [31]. The condition improves in less than 25% of patients; remission of the main symptoms occurs only in 6% of patients; continued deterioration is observed in 1/3 of cases [29].

The presented clinical example is complex not only in terms of interpreting manifestations of the motor phenomenon as a whole but also in terms of the degree of a patient's concern with his own problem. The patient uses antagonist gestures that are now considered to be a characteristic sign of dystonia [33, 34]. Neither the medical history nor the patient complaints have had indications for the characteristic symptoms of hysterical disorder, such as voice and speech disorders [35], polymorphic psychotic disorders, multiple somatic complaints, pains in the affected limb during passive motion, and other paroxysmal conditions. However, the patient has other, very typical of psychogenic dystonia, syndrome features: sudden onset of the disease with rest dystonia, fixed muscle spasm, and the absence of dynamics of the movement pattern typical of dystonia [2, 29, 31, 36, 37]. In the described clinical case, there is a negative dissociation between the presented complaints and the clinical manifestations, which is very typical of PMD. Indeed, the patient's main and only complaint is his motor "defect" in the right hand-involuntary clenching of the hand; in this case, on examination, a number of features are presented (demonstrated) in the manifestation of this phenomenon, e.g., dramatic crooking of the fingers like a "clawed paw". Another motor phenomenon presented by the patient during writing is elements of focal hand dystonia (writer's cramp). It should be noted that all patients with idiopathic writer's cramp have a certain ritual-preparation before starting to write, including a characteristic dystonic pose of the hand and fingers and apparent dynamics of the hyperkinetic movement pattern; in this case, an inevitable change in the handwriting feature occurs during the disease [12]. In the presented case, the manifestations typical of classical writer's cramp and the staged development of dystonia are absent.

The patient "developed" a whole system of measures to reduce "involuntary" clenching of the hand: the use of leather gloves or "finger expanders"; holding right fingures 3 and 4 crossed; holding an object with a hand being in the supine position; extending the hand during writing by pressing it to the Table 1. Of particular attention are phenomena such as the crossing of fingures 3 and 4 and the fanciful position of the hand in the supine position when holding an object. Each of these phenomena is extremely inconvenient in terms of both its implementation and holding of the hand in this position. In the presence of these movement hand changes that significantly affect his living activities, the patient refuses to consult a psychiatrist and proposed treatment. Noteworthy that in the presence of demonstrated forced "involuntary" postures of the hand, the neurological status has no signs of increased (spastic or plastic) muscle tone in the hand and arm; there is also no asymmetry of tendon reflexes, atrophies of the hand and forearm muscles, and pain syndrome. According to the laboratory and instrumental examination, the patient has no changes indicating involvement of the central or peripheral nervous system.

In the presented case, the complex of the described features enables the diagnosis of a psychogenic movement disorder in the form of fixed dystonia not accompanied by pain.

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