

Functional writer's cramp as psychogenic focal dystonia

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We present a description of a functional writer's cramp case. The disease manifested as pain and tension in the right hand when writing; thus, we suspected kinesigenic dystonia in the form of writer's cramp. However, the motor pattern and the presence of additional manifestations made it possible to assume the neurotic nature of hyperkinesia. A psychiatrist diagnosed a combined conversion motor and undifferentiated somatoform disorder as a part of personality dynamics of the dramatic cluster at the involutionary age. Treatment included cognitive-behavioral therapy and periciazine administration with a positive effect. We discuss the aspects of hyperkinesia and mental status, which help differentiate the kinesigenic form of dystonia (writer's cramp) and functional movement disorder.

Keywords: movement disorders; dystonia; writer's cramp; kinesigenic dystonia; functional movement disorder; conversion disorder.

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For reference: Tolmacheva VA, Yuzbashyan PG, Shishorin RM, et al. Functional writer's cramp as psychogenic focal dystonia. *Nevrologiya, neiropsikhiatriya, psikhosomatika = Neurology, Neuropsychiatry, Psychosomatics*. 2022;14(1):115–121. DOI: 10.14412/2074-2711-2022-1-115-121

Motor disorders are typical of multiple neurological diseases. A substantial proportion of those (about 20%) belong to dystonic hyperkinesia. [1, 2]. Dystonia started to be perceived as a neurological disease in the middle of the past century, when D. Marsden [3] and S. Fahn [4] provided definition of dystonia and created the classification of dystonic syndromes. Arguments confirming organic/neurological nature of focal dystonia included the fact that in some cases generalized dystonia with early onset can be manifested as a focal form. Therefore, focal forms can represent early stage of the same disease that progresses later on. Arguments in favor of this point of view also include the fact that generalized dystonia with late onset, like focal dystonia, does not progress, is not inherited and is comparable with focal one in terms of the age of onset. This point of view was additionally supported by new data on the existence of hereditary forms of the disease (including discovery of genes associated with dystonia) [5].

Writer's cramp is one of the most prevalent types of upper arm kinesio-specific dystonias. The disease pathophysiology is not well-known, although available data indicates disorders of inhibitory mechanisms on the cortical, brainstem and spinal cord levels, as well as abnormalities of the sensor systems and cortical flexibility in genetically predisposed individuals [6–14]. Moreover, features of hand movement biomechanics play some role in the pathophysiology of writer's spasm [15].

Supposedly, activities involving prolonged writing are important factors contributing to the disease development [16, 17].

The strict relation with specific type of movement (writing) is lost during disease progression, and dystonia can occur during other activities, which results in significant reduction of the quality of life of these patients. Local injections of botulinum toxin are currently considered to be most effective treatment [18–20]. Its disadvantage is the necessity to perform repeated injections approximately every 3 months. Rehabilitation approaches based

on sensor and motor trainings are also successfully used to prolong therapeutic effect [21, 22].

Dystonia is often difficult to diagnose. It is not always easy to identify dystonic hyperkinesia, especially in cases of mild, sub-clinical or atypical forms [23, 24]. Functional forms of hyperkinesia in which cervical dystonia masks a psychic disorder are also well-known [25].

Relatively little is known about functional writer's spasm, so we provide our clinical case description.

Patient Sh., 56 years old, primary care physician and functional diagnostics specialist.

Anamnesis morbi: The patient considers herself ill since the age of 28, when after a maternity leave, she started to experience discomfort during various kinds of paperwork — writing case histories, medical notes and reports. She found out that her right arm could not be completely extended and remained half bent for a long time after she finished work. She felt contraction in the hand and unintentional index finger abduction which interfered with work; she «had difficulties holding a pen». She also complained of dull pain in the wrist which increased during active movements. She noted that she did not feel any discomfort or any movement limitation during prolonged rest, i.e. on vacation. Within the first two years the pain resolved, although episodic motor hand disorders remained. The patient state was relatively stable in the period from 30 to 50 years old: she felt tension, unintentional extension of the index finger during prolonged writing, but there was no pain syndrome. The patient continued working as a doctor, and the remaining symptoms did not have any influence on the occupational activity.

Menopause started at the age of 48, it was accompanied by hot flushes, irritability, easy crying. At the age of 50 after vaccination against influenza and increased workload on the hand at work, as well as disagreement with her boss, she registered worsening represented by diffuse feeling of heaviness in the hand. During the next

year she suffered from pain syndrome in the right hand during writing, she experienced difficulties with moving right fingers and started to have significant weakness. Sleeping disorders followed, she could not fall asleep for a long time and was looking for a comfortable position to decrease the pain. It was then that she sought medical assistance from a local neurologist and was diagnosed with «segmentary right hand dystonia». The patient received physiotherapy, botulinum therapy in the right forearm and hand without significant effect. She was referred to the Neurological Disorders Clinic in Sechenov University for evaluation and treatment.

Neurological status: the patient complains of tension and pain in the first three right fingers, which leads to unintentional movements of the right hand and significantly interferes with writing. The clinical examination revealed hyperkinetic disorders of the right upper limb; unintentional flexion of the hand with extension of the index. Arm extension leads to unintentional movements in the right hand fingers and incomplete flexion of the wrist with finger extension. Similar movements appear when the patient is asked to hold the arm in any position. The movements of the right hand fingers are slowed and clumsy on evaluation. Tenderness in the index finger spreading to the hand and wrist is noted, but not in all diagnostic tests. Movement patterns are variable when the same tests are repeated after time intervals. At rest episodic unintentional minor finger movement can be seen.

No pathological changes are observed on encephalography and brain magnetic resonance tomography.

Neurologist's opinion: functional writer's spasm.

A psychiatrist's consultation was recommended considering the characteristics of movement patterns of hyperkinesia and anamnesis data. The patient provided informed consent for consultation.

Mental status: no known hereditary overt psychosis. The patient's mother (87 years old) also worked as a general practitioner until retirement. The patient's behavior in the family — strict, autocratic, capable of punishing for disobedience, frequently raises her voice. After 35 years old she started to suffer from pain in her right hand, neurologists diagnosed «writer's spasm», the patient has been suffering periodical pain relapses until recently and takes painkiller drugs. The patient grew up as a communicative child and had difficulties being alone. She liked to draw attention to herself, wanted to be on stage since early childhood, sang and read rhymes on family parties on her own initiative. She chose a number of «trustworthy» friends when she was a part of the group of children. She acted as a leader among them, commanded and actively involved them in her activities. She started to go to take dancing and singing lessons since she was 4, and took piano lessons since the age of 6. The patient attended children's artistic clubs and frequently went on tour with them. She considered herself a better student than her peers. She went to school at the age of 6 and adapted quite well. She was an excellent student and thoroughly performed all the tasks. She liked to speak in front of the class, did not feel embarrassed and was always confident about her knowledge. She actively and happily participated in extracurricular activities. Since she was in the 6-th grade, she started studying chemistry, participated in academic competitions, prepared for studying in medical university.

Since childhood the patient suffered from headaches, which she felt as girdle-type pressure, especially in the frontal area, that increased in the evening or after physical/emotional tension. She did not tell anything about pain to adults, they did not have any influence on her activity and mood.

Her grandfather died when she was 10, she had a hard time, was depressed, felt frustration, had sleeping disorders (difficulties in

falling asleep). After the funeral she saw her grandfather's shadow moving across the room when she was alone; this resulted in her being scared, she cried and tried to pray. After that, she had troubled sleep, with several wake-ups. Her state recovered without assistance.

She had regular painful menses since the age of 12. She had hard time during premenstrual period: every time she started feeling weakness, fatigue, was tearful and irritated for a few days before menses.

The patient graduated from school at the age of 17, when she entered a medical university in Moscow. Despite the fact that earlier she did not leave parents for a long time, she moved with ease, liked the atmosphere of a big city and quickly adapted to students' life. However, she could not get in contact with her peers — dorm neighbors, she blamed them out loud for «wrong» life style, which resulted in conflicts and led to frequent changes of rooms where she lived. She received excellent marks, liked the chosen occupation, spent all her free time preparing for lessons, and later started to work as a hospital attendant. She was sure she had a «gift» for being a doctor, boasted of her ability to understand patients, and thought that while being a hospital attendant, she possessed knowledge and skills equal to those of surgeons. She intentionally did not keep distance with patients, tried to gain their trust, to make good impression.

At the age of 20 she had hard time during the divorce of her parents, felt depressed, had sleeping problems. She felt anger, took offence at her father who, in her opinion, destroyed the family. A few weeks after her parents' divorce she started to feel irregular heartbeat, which seemed to be like temporary «arrest» of cardiac rhythm followed by tachycardia. This persisted for about half a year, then it gradually reduced, however, episodes of irregular rhythm continued later in cases of jitteriness.

She married her peer at the age of 23. She had normal births at the age of 24, 27 and 35 years old. She mentioned no depressed mood. Her family life was not very good, right after the first delivery she started to quarrel with her husband, accused him of little help and small salary. She frequently suspected unfaithfulness, tried to confirm her suspicions, followed the husband, checked his clothes, and called his colleagues. When she eventually confirmed his adultery, she found his lover, and this resulted in an open scandal, trying to return her husband home. When the patient was 33, her husband initiated divorce. During the divorce period she was tearful, irritated, frequently took it out on children; she claimed to feel depressed, did not feel happy regarding the activities she previously enjoyed, it was difficult for her to contact with people. However, she continued communication with men and got married once again a year later. The relationship with the second husband was not good either, and after a few years she initiated divorce. Now the patient lives with her boyfriend.

After the patient graduated from the university and completed her residency, she worked as a general practitioner for her whole life, both in state hospitals and in private clinics. She took a number of postgraduate courses and became an ultrasound specialist. She remained in one place for a long time and changed her workplace only in cases of profitable offers. At work she took part in various activities: organized office parties, talent groups, in which she always was a leader and key figure.

The patient looks younger than her age, she wears fancy outfit and lots of jewelry. She has a lot of make-up, and a complex hairstyle. She is communicable and welcoming. No schizoid disorders found. The patient complains of pain and movement disorders in her right hand. She says that she feels nagging pain in the fingers during armload. From time to time, it spreads to the rest of the hand both

from the palmar and dorsal sides in a «glove-like» type. This feeling switches to «unbearable» burning – «as if I scalded my hand». According to her words, these feelings do not allow her to complete movements in the joint and result in unintentional extension of the index. She describes her complaints in detail, with emotional coloring. According to the patient, the pain regularly becomes «horrible», «agonizing» and she cannot distract her mind from it. She also dramatically describes movement disorders, despite their small amplitude on examination; she says that she is unable to perform not only her professional tasks, but also everyday activities (in the meantime the pain does not interfere with recreational activities, such as amateur performances, playing the music). According to her, she started to «use her disease to avoid overtime work» by claiming to have severe pain and «writer's spasm», she declines additional work and many everyday activities that require armload are fulfilled by the boyfriend.

She describes herself as an emotional person with trends to frequent mood changes. She confirms frequent manipulations in her relationships with partners, justifying her behavior by her health issues and her children's welfare. In the past she reacted to serious stress, i.e. work conflicts, not only by short-term depression and irritation but also by physical malady (pain, mostly in hands; numbness of hands and feet of «glove and sock» type; heart arrhythmia, feeling of «lump in the throat»). She did not consult specialists regarding this issue, because she understood the transient nature of these feelings, which quickly resolved until recently. She is not afraid that pain and hand movement disorders can be caused by some undiagnosed severe neurologic disease. She confesses that several times she searched for «more appropriate diagnosis» in neurological literature. She imagined herself completely immobilized as a result of possible progression of pathology.

A psychiatrist's diagnosis: complex conversion motor (F44.4) and undifferentiated somatoform disorder (F45.1) within personality dynamics of personality disorder of dramatic cluster (F60.4) at the involutional age.

The patient received a course of cognitive-behavioral therapy (five sessions), targeted at catastrophizing phenomenon in terms of health-based anxiety, as well as dysfunctional personalized patterns within pathocharacterological structure of dramatic cluster. She also received drops of periciazine in addition to psychotherapy (titrated up to 10 mg/day, before nighttime) accompanied by blood pressure control. The therapy resulted in complete reduction of both conversion and somatoform symptoms within a month without any relapse within next six months.

Discussion

In this case the type of hyperkinetic disorder is different from typical kinesio-specific dystonia, such as writer's spasm. According to the patient, the disease was diagnosed when the patient was young, however, no medical documentation was provided. The disease course was favorable for a long time, tension and rigidity in the forearm did not cause any inconvenience and did not result in changing occupation which is not characteristic of patients with typical writer's spasm. The period before formation of hyperkinesia which resulted in inability to write, lasted for more than 20 years. Such a long «prodromal» period is also not typical of the development of writer's spasm. Neurologic examination revealed unintentional hand flexure with index extension during writing, which reminds of typical pattern in focal muscular hand dystonia. However, the writing disorder per se is not isolated. The patient complains not only that it is «difficult to hold a

pen», but she also has difficulties with other actions, for example, flexure and unclenching of hand, playing with fingers, difficulties in abduction and adduction of fingers etc. Thus, dystonia is observed not only during writing.

However, the same tests showed different results when performed several times with time intervals, which serves as one of the «positive» signs of conversion nature of psychoneurological disorders [26, 27]. Moreover, the patient performs diagnostic tests slowly and clumsily, and complains of rapidly evolving weakness in the hand. Muscular weakness and slowness in the affected limb are not typical of dystonia [28]. On the contrary, typical writer's spasm does not cause weakness, but leads to extra tension in the muscle, due to which the patient has to terminate respective activity. This tension in some cases can result in pain.

It is important to notice that in this observation pain is the dominant and primary symptom, while tension and motor disorders appear later. In typical cases of classic dystonia (writer's spasm) pain appears after the feeling of muscle tension [29]. Periodic twitches of fingers that occur in the patient at rest are also unrelated to the diagnosis «writer's spasm».

Besides, patients with focal muscular dystonia typically use various corrective gestures or devices to cope with compulsory dystonic movements [30]. This patient did not use any kind of assisting methods.

In this case during stressful situations or when unpleasant job needs to be done (mainly related to professional activity) these sensations significantly increase, and she does not have any movement limitations when she feels emotional comfort (during recreational activity, playing the music), which is also a sign of psychosomatic disease.

At the time of examination, the patient's mental state can be categorized as manifestation of comorbid conversion and somatoform disorder in a person with dramatic cluster personality disorder. This serves as the substantiation of psychogenic dystonia syndrome, which matches the criteria of incongruity [28, 31]; the signs of «dystonia» in the discussed case are not in line with the classic signs of writer's spasm.

The conversional nature of hyperkinetic symptoms in addition to atypical «movement pattern» is confirmed by their episodic, versatile and situational appearance, their relation to certain, subjectively important external stress factors, first of all, conflicts at work, during the patient's habitual acts «in public».

Hysterical nature of pseudoneurological disorders is also confirmed by combined motor and sensor dysfunction topographically limited by hand region [32]. In this case we refer to the coupled functional motor (dissociative in terms of modern classifications – DSM-5 and ICD-11) and pain disorder, categorized as somatoform from the formal point of view of modern systematics (somatic symptom disorder in DSM-5 or bodily distress disorder in ICD-11). Pain in the hand can be categorized as hysterical algia in terms of psychopathological classification (hysteroalgia) in Russian articles [27, 33], mostly in accordance with their dramatized and vivid descriptive characteristics, changing of intensity and localization in the hand region.

The patient behavior during the period of disease corresponded to hysterohypochondria: she is manipulative and creates a special «safe» regimen, that includes declining from additional workload and delegation of «unpleasant duties» to the family members [34], which also confirms hypochondriac anxiety. Anxiety regarding health issues is dramatized: despite reassurance of health care professionals, the patient does not exclude the pos-

sibility of a severe neurological disease and applies the symptoms from medical literature to herself, creating vivid horrifying images of disability due to irreversible damage to the limb.

We should note the following aspects while switching from syndrome categorization of psychic pathology under discussion to nosological diagnosis. Disease course correlates with the trajectory of personality disorders. Motor disorders that were episodic and quickly resolving during the disease onset (at the age of 20), at involutional age after menopause became chronic, constant, «habitual» stereotypic movements associated with a number of other functional (conversion and somatoform) symptoms both in the hand region and outside of it (clavus hystericus and others) [35].

This nosological classification is confirmed by family history («hysterical copying of mother's symptoms», who had writer's spasm), static and dynamic signs of hysterical personality disorder (dramatic cluster B), including dissociative and somatized hysteroconversion reactions in the past; possibility to exclude other psychiatric/neurological diseases during differential diagnostics (schizophrenia, affective pathology, organic disorders of the central nervous system).

The diagnosis confirms efficacy of cognitive behavioral psychotherapy, which addressed catastrophizing as a part of health-related anxiety, and dysfunctional personality scheme within pathocharacterological structure of dramatic cluster [36]. Periciazine that we used could be effective not only in personality disorders but in some cases in their «derivates» — conversion-dissociative (hysterical) phenomena [37].

Although patients with psychogenic hyperkinetic disorders can also be given botulinum therapy, in this case it was decided

not to use this approach. Since botulinum therapy was previously used in the patient without significant effect, we could hardly expect positive effect right after the injection («on the needle») which in such cases is qualified as confirmation of functional nature of the disease (placebo effect) [38].

Clinical analysis of hyperkinetic disorder in the hand, incompatibility with classic dystonia criteria do not allow us to categorize it as kinesio-specific dystonia (writer's spasm) [39]. In this situation as well as in other cases when dystonic hyperkinesia has uncharacteristic pattern, is accompanied by other neurological symptoms, absence of response or, on the contrary, immediate response to botulinum therapy, consultation of a psychiatrist is necessary for evaluation of psychic status and personal characteristics of the patient. Modern approaches to treatment of functional dystonia are based on common principles of management of patients with psychogenic diseases. Cognitive-behavioral therapy [40], psychodynamic psychotherapy and various physical rehabilitation programs [41] are most frequently used. Unfortunately, the prognosis for patients with functional dystonia is generally unfavorable [42]; frequently the patients do not feel improvement even after several courses of treatment [38, 43], however, the severity of symptoms can vary, and short-term remissions are possible [44]. Despite poor or unstable response to treatment, symptoms may improve in some patients, and sometimes treatment results in complete remission [43, 45].

Taken into consideration the absence of the «gold standard» of management of such patients, treatment should be individualized, adapted to the type of physical symptoms and the level of accompanying psychic pathology, and should include physical rehabilitation, psychotherapy and treatment of comorbidities [46].

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Received/Reviewed/Accepted
27.10.2021/11.12.2021/17.12.2021

Conflict of Interest Statement

The investigation has not been sponsored. There are no conflicts of interest. The authors are solely responsible for submitting the final version of the manuscript for publication. All the authors have participated in developing the concept of the article and in writing the manuscript. The final version of the manuscript has been approved by all the authors.

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