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Is there a specific psychiatric background or personality profile in functional dystonia?

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ABSTRACT

Objective: The aim of this cross-sectional study was to identify if there was a specific difference between patients with functional dystonia (DysF) and those with adult-onset, isolated idiopathic ("primary") dystonia (DysP) in terms of psychiatric disorders, psychological stressor, dissociation correlates, and personality traits.
Methods: Thirty-nine clinically definite DysF and 30 DysP patients matched by age, gender and dystonia distribution underwent psychiatric interview based on DSM-5 criteria and additional testings for global cognitive and psychiatric functions (Mini-Mental State Examination, Hamilton Depression and Hamilton Anxiety Rating Scale, Apathy Scale, Somatoform Dissociation Questionnaire-20, Dissociative Experiences Scale II, and the five-dimensional Revised Neuroticism-Extroversion-Openness Personality Inventory).
Results: Almost half of our DysF patients had prior psychiatric treatment, which was significantly more frequent when compared to DysP. Patients with DysF in comparison to DysP also had considerably more frequent preceding stress, higher apathy, dissociative and somatoform scores, as well as significantly higher rate of *la belle indifférence* sign. This sign, stress before dystonia and prior psychiatric disorder independently predicted having DysF. Some of psychiatric disorders (i.e. substance-related disorders, schizophrenia, adjustment disorder, borderline personality disorder, post-traumatic stress disorder, psychotic depression, delusional disorder) were exclusively present among DysF patients. DysF compared to DysP patients had lower scores for both extroversion and openness to experiences.
Conclusion: Our data found different pattern of psychiatric comorbidity and personality traits between DysF and DysP patients, including a higher prevalence of psychological stressor and dissociative correlates, indicating at least a partial role of psychological mechanisms in the pathogenesis of DysF.

1. Introduction

Nosology and assessment of functional (psychogenic) neurological disorders (FND) have a long history, and are continuously under debate [1]. The key issue is the absence of pathophysiological understanding with numerous etiological theories about functional disorders that could be put into the frame of one of two main concepts: "brain" or "mind" [2]. Traditionally, the term "conversion disorder" (the closest equivalent to FND based on the DSM-IV) has described pseudoneurological symptoms that were not attributable to nervous system disease or to a feigning, but were considered to be associated to psychological factors [3].

Increasing research interest in FND that revealed complex pathophysiological interplay among different features like specific biological vulnerability [4], personality characteristics [5], early childhood trauma [6], social and family modeling [7], and physical trauma [8] moved the focus away from psychological stress. Currently, the presence of psychological factors preceding FND/FMD is under debate, since the DSM-5 criteria for conversion disorder (functional neurological symptom disorder) [9,10] do not require "psychological stressor" as a diagnostic criterion [11]. Furthermore, substantial proportion of patient with functional movement disorders (FMD) were within normal score ranges when relevant psychological questionnaires/tests were applied [12]. However, it is still important to identify a wide spectrum

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of various psychiatric comorbidities, such as depression, anxiety and personality disorders, frequently reported in patients with FND/FMD [13–15], since they are associated with poor outcome [14].

Majority of data cover FMD as a group [6,13,14], and little is known about specific underlying pathogenesis of functional dystonia (DysF) [16,17], probably the most severe form of all FMD, often accompanied by complex sensory [16] and psychiatric feautres [18]. Despite the fact that isolated ("primary") dystonia (DysP) has traditionally been considered as an exclusively motor disorder, a growing body of evidence reported the presence of neuropsychiatric disturbances, especially anxiety and depression, even before the onset of dystonic movements [19,20]. The relationship of psychiatric comorbidity to the pathophysiology of DysP is unknown [21]. Such association may hypothetically be even more relevant for DytF.

Therefore, the aim of this study was to identify if there was a specific psychiatric/psychological background of patients with DysF in comparison to those with DysP, in terms of psychiatric disorders, psychological stressors preceding dystonia, dissociative correlates, and personality traits.

2. Patients and methods

In this cross-sectional study, 39 patients with dystonia fulfilling criteria of "clinically documented" FMD proposed by Gupta and Lang [22] were included in further analysis. In addition to acute mode of onset and fixed posture of extremity and neck, other features of inconsistency or incongruence with organic dystonia included: a) variability in the performance of involuntary movements, including amelioration of dystonia during distraction, b) unusual age at onset for; c) persisting unilateral or asymmetric symptoms (e.g., bilateral but asymmetric or unilateral orbicularis oculi spasm with contralateral frontalis overactivity or lower face dystonia), d) history of spontaneous amelioration or remission of abnormal movements, e) severe and early pain, f) unexpected response to botulinum toxin injections, and/or suggestions. Standard investigations for secondary dystonia [23] were normal in all patients, including brain MRI finding. Genetic tests for mutations in the DYT1 and DYT6 gene were negative in all patients, while in cases resembling dystonia-myoclonus phenotype mutations in DYT11 gene was excluded. DysF patients were matched by age, gender, and dystonia distribution with 30 patients with DysP (Table 1). After signing informed consent, all patients underwent careful clinical examination and completed questionnaire regarding various demographic and clinical features (including trigger for dystonia, pain, etc.). All patients were examined by experienced movement disorders specialists (VSK, IP, MS, NDM). The disease severity was assessed by the Unified Dystonia Rating Scale [24], Burke-Fahn-Marsden Dystonia Rating Scale [25], and the Psychogenic Movement Disorders Scale [26].

Thorough psychiatric assessment included psychiatric interview based on DSM-5 criteria [9] (DP, AP) and additional tests for global cognitive and psychiatric functions: the Mini-Mental State Examination (MMSE) [27], Hamilton Depression Rating Scale (HDRS) [28], Hamilton Anxiety Rating Scale (HARS) [29], Apathy Scale (AS) [30], Somatoform Dissociation Questionnaire (SDQ-20) [31], and the Dissociative Experiences Scale II (DES-II) [32]. The five-dimensional Revised Neuriticism-Extroversion-Openness Personality Inventory (NEO-PI-R) [33] was used for quantification of personality traits. Medical records and psychiatric interviews were used as a source for psychiatric diagnosis before dystonia onset. *La belle indifférence* sign was assessed according to the traditional description of "relative lack of concern about the nature or implications of symptoms" [3,34]. This sign was assessed during psychiatric interview, and then further observed during regular controls.

The study was approved by the Ethical Committee of the School of Medicine, University of Belgrade (Serbia).

In cases of data with non-normal distribution among the continuous variables, the Mann-Whitney U test was used to investigate the differences between the two groups. To explore differences in discontinuous variables, the Chi^2-test and McNemar test were applied. To identify the main psychiatric predictors of having DysF vs. DysP, logistic regression analysis was conducted, with dystonia type as the outcome variable, and with general psychiatric variables as predictors.

Table 1
Demographic and clinical features of patients with functional and primary dystonia.

Variable	Functional dystonia	Primary dystonia	p
Age (years)[a]	46.5 ± 14.7 (17–71)	44.0 ± 12.9 (19–62)	n.s.
Sex (m/f)[b]	10/29 (34%)	5/25 (20%)	n.s.
Education (years)[a]	11.3 ± 1.5 (8–14)	12.1 ± 1.2 (11–16)	n.s.
Handedness (right)[b]	35 (89.7%)	28 (93.3%)	n.s.
Hereditary dystonia[b]	1 (2.6%)	6 (20%)	0.038
Age at onset (years)[a]	41.9 ± 13.4 (19–65)	27.1 ± 17.0 (2–55)	0.000
Disease duration (years)[a]	4.6 ± 4.2 (0–18)	16.2 ± 12.2 (1–44)	0.000
Trigger[b]	21 (53.8%)	6 (20%)	0.004
Trigger at dystonia site[b]	6 (15.4%)	5 (16.7%)	n.s.
Trigger-dystonia latency (months)[a]	2.6 ± 4.7 (0 − 12)	3.0 ± 2.7 (1–6)	n.s.
BTX treatment[b]	17 (43.6%)	28 (93.3%)	0.000
Efficacy of BTX (%)[a]	44.7 ± 23.0 (0 − 100)	49.3 ± 18.8 (20–80)	n.s.
Sensory trick[b]	0 (0%)	8 (26.7%)	0.001
Pain[b]	29 (74.4%)	13 (43.3%)	0.009
Severe pain[b]	16 (55.2%)	6 (46.2%)	n.s.
MMSE[a]	28.3 ± 1.56 (25–30)	29.0 ± 1.3 (26–30)	0.078
BFMS[a]	11.7 ± 8.6 (2–34)	16.9 ± 18.2 (0–65)	n.s.
UDRS[a]	11.2 ± 7.9 (2 − 30)	22.9 ± 22.8 (5–84)	0.020
PMD total phenomenology score[a]	10.8 ± 4.8 (4–33)	/	/
PMD total functional score[a]	7.8 ± 3.6 (0–12)	/	/
PMD total score[a]	18.9 ± 7.0 (4–32)	/	/

n.s. = not significant; BTX = botulinum toxin; MMSE = Mini-Mental State Examination; BFMS = Burke-Fahn-Marsden Dystonia Rating Scale; UDRS = Unified Dystonia Rating Scale; PMD = Psychogenic Movement Disorders Scale.
[a] Values presented as means ± SDs, with a range in parenthesis.
[b] Values presented as numbers of patients with percentage in parenthesis.

3. Results

General psychiatric features of both groups are presented in Table 2.

Table 2
General psychiatric variables in functional and primary dystonia.

Variable	Functional dystonia	Primary dystonia	p
Stress before dystonia[b]	28 (71.8%)	2 (6.8%)	0.000
Type of stress:			
Interpersonal[b]	7 (26.9%)	2 (100%)	n.s.
Other[b]	19 (73.1%)	0 (0%)	
Psychiatric heredity[b]	2 (5.6%)	2 (6.7%)	n.s.
Psychiatric disorder			
Prior[b]	22 (61.1%)	7 (23.3%)	0.002
Current[b]	26 (72.2%)	12 (40.0%)	0.008
Prior psychiatric treatment[b]	17 (47.2%)	5 (16.7%)	0.009
Prior AD therapy[b]	17 (47.2%)	5 (16.7%)	0.009
Apathy scale[a]	16.8 ± 11.6 (0–40)	10.9 ± 8.5 (0–28)	0.037
HDRS[a]	15.7 ± 10.3 (0 − 33)	10.9 ± 8.5 (0–28)	0.072
HARS[a]	13.4 ± 10.4 (0–39)	9.9 ± 7.3 (0–25)	n.s.
DES-II[a]	4.3 ± 6.9 (0–25)	0 (0%)	0.000
SDQ-20[a]	29.3 ± 10.2 (20–58)	20.2 ± 1 (20–25)	0.000
La belle indifférence[b]	26 (70.3%)	5 (16.7%)	0.000

n.s. = not significant; HDRS = Hamilton Depression Rating Scale; HARS = Hamilton Anxiety Rating Scale; DES-II = Dissociative Experience Scale II; SDQ-20 = Somatoform Dissociation Questionnaire.
[a] Values presented as means ± SDs, with a range in parenthesis.
[b] Values presented as numbers of patients with percentage in parenthesis.