Single Case – General Neurology

Chronic Functional Tremor: Positive Signs in the Management, Including Mirror Therapy

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Keywords

Functional tremor · Functional neurological symptom disorder · Biopsychosocial treatment · Mirror therapy

Abstract

Functional tremor (FT) is a common functional neurological symptom disorder (FNSD) and difficult to treat. Diagnosis is often delayed in FNSD, which reinforces maladaptation and chronicity. The presented case, who had suffered from FT for 2 years, demonstrates the value of positive neurological symptoms in the diagnosis and treatment of FNSD, even in a chronic case. The patient improved well during integrated inpatient rehabilitation, which included mirror therapy (MT). He had complete suppressibility of the tremor during distraction, which was shown to him, and reversibility of the FT was emphasized. Due to the suppressibility when focusing on the contralateral arm, we installed MT as part of a combined inpatient neurological-psychotherapeutic rehabilitation treatment lasting 8 weeks. During treatment, some strong emotional themes came up. At the end of rehabilitation, the tremor almost completely subsided. Possible pathomechanisms are discussed. Future studies are recommended for determining the effectiveness of MT in FNSD with one-sided symptoms.

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Introduction

Functional tremor (FT) is a common functional neurological symptom disorder (FNSD) [1, 2]. The etiology is not well understood [3]. The Diagnostic and Statistical Manual of Mental Disorders, fifth edition (DSM-5), omitted psychological factors as sine qua non for the diagnosis for FNSD [4], and the International Classification of Diseases, eleventh edition (ICD-11, <https://icd.who.int/browse11/l-m/en>), will go the same way. In particular, DSM-5 emphasizes positive neurological signs, which have good sensitivity and specificity [5]. These can enhance the diagnostic process considerably. The delay until definite diagnosis is often long in FNSD, in dissociative seizures up to 7 years [6]; and early diagnosis and treatment beginning as well as treatment satisfaction are important prognostic factors in FNSD [7]. Furthermore, demonstrating these signs to the patient is helpful in emphasizing the functional and reversible nature of the disorder. Though there is some debate around differential diagnosis, there is limited evidence for treatment [8]. For complex cases, multimodal inpatient treatment considering functional as well as psychotherapeutic approaches is advocated [9, 10]. Also, when communicating the diagnosis and at the beginning of treatment, a joint neurological-psychotherapeutic approach seems promising [11]. In the presented patient, inpatient rehabilitation treatment consisted of a combined psychotherapeutic and functional neurological treatment including mirror therapy (MT). MT uses visual feedback from the unaffected arm by watching its movement in the mirror, and it is effective in stroke patients [12]. Although functional treatment modules receive attention in FNSD [13], we are not aware that MT has specifically been considered in FT or FNSD. The case is also unusual as symptoms had been present for 2 years, i.e. chronic, and he improved over an 8-week inpatient treatment.

Case Presentation

A 64-year-old married man had worked as a computer scientist for decades. He had been suffering from tremor of the right arm for 2 years. It was a resting and action tremor, which was completely suppressible with distraction, e.g. when focusing on the contralateral arm (online suppl. Video 1; see www.karger.com/doi/10.1159/000507567 for all online suppl. material). The tremor was incompatible with other neurological conditions, and therefore fulfilled criteria for FNSD according to DSM-5. There were no other abnormalities of the neurological examination. Nevertheless, previously he had had an extensive outpatient workup including a dopamine transporter scan and treatment with L-DOPA without any success. After a (quite typical) diagnostic odyssey, he was eventually diagnosed with FT and received inpatient psychosomatic treatment which focused on psychotherapeutic issues. There was no improvement of the FT. The patient had problems with fine motor skills like buttoning, handwriting, or using the keyboard, and he had been off work for 16 months. There had been no previous psychiatric history. He then entered our integrative psychotherapeutic-neurological inpatient rehabilitation program consisting of physiotherapy, occupational therapy, individual and group psychotherapies, art therapy, etc. [details in 9]. Occupational therapy used manual therapy to relax muscles and MT due to the suppressibility when focusing on the other arm (online suppl. Video 2 and 3). And we thought that this therapy might enhance integrative processes of the left cerebral hemisphere through visual response via the mirror (suggesting the right arm performs tasks instead of the left). He trained MT by himself on a daily basis for 15–20 min after initial instruction by the therapist. As recommended in the literature [1, 8], we demonstrated suppressibility of the tremor to the patient himself and explained the

functional nature and potential reversibility. During individual psychotherapy (50 min per week) and group psychotherapy (three times 50 min per week), he was increasingly able to open up. When asked about subjective theories of the disorder, he mentioned (orthopedic) problems with the neck as well as problems at work: Because of restructuring, he had less to do and he had obviously suffered from “boreout.” This was a problem for this conscientious man who developed (psychodynamically) an “uneasy right hand.” Furthermore, during the second half of treatment, he showed deep affection and emotional pain: his wife had been suffering from pulmonary fibrosis for 6 years being on continuous oxygen therapy, and she had been transplant-listed (with little prospect to receive a transplant). He himself had completely stopped all social activities.

With emotional relief and functional therapy over 8 weeks, the tremor was substantially reduced (online suppl. Video 4). He performed fine motor tasks more easily with near normal manual speed, e.g. when using a keyboard or when writing. Furthermore, hand-hand and hand-eye coordination was quicker and target-aimed, e.g. when collecting coins or opening small buttons. Improvement was sustained according to a 2-month follow-up.

Discussion and Conclusions

FNSD is very common in neurological practice, and prognosis is poor in more than 50% of cases. This also includes high socioeconomic burden. Etiological concepts of the disorder shift from psychoanalytic thinking to a more integrated biopsychosocial approach, which also influences treatment concepts [3]. There are a couple of diagnostic and treatment-related issues to be learned from this presented case, which are detailed below. *Nota bene*: The diagnostic process is itself the “door-opener” to treatment, in particular in functional and somatoform disorders, i.e. diagnostic and treatment processes are tightly interwoven [14]. Here, psychoeducational approaches including demonstration of the signs to the patient and potential reversibility are important [8]. This involves issues of the therapeutic relationship, countertransference etc., which have to be considered in the medical management of these, at times considered difficult, patients.

1 Clinical diagnosis of FNSD/FT can be accomplished rapidly with positive functional neurological signs [8]. Typically, these patients have long diagnostic odysseys with foci on excluding other somatic diseases, which is costly and perpetuate the disease process. In effect, the role of the neurologist is strengthened not only to exclude other somatic disorders, but to use “positive signs” to make the diagnosis. In this case, it took more than 1 year to establish the correct diagnosis, although with careful examination and focus on positive signs for FNSD, it could have been made earlier.

2 Demonstrating these positive signs to the patient is a powerful tool to build up a trusting and respectful therapeutic relationship as well as explaining the functional and therefore potentially reversible pattern of the disorder [8]. This also strengthens the *therapeutic role* of the neurologist.

3 Subjective personal theories of the disorder are helpful and nurture therapeutic relationship in the context of an integrative biopsychosocial attitude and treatment [15].

4 Disintegrative cerebral processes have long been discussed as relevant in FNSD [16, 17], and there is one report on entrainment therapy in FT [1]. For this patient, we used MT, which we are also using for functional hemiparetic symptoms in our Institution [18]. One might consider the integrative processes of this therapy using both cerebral hemispheres, i.e. when performing tasks with the unaffected left hand, the patient perceives the right arm,

therefore making use of or inducing perceptive processes of the left cerebral hemisphere; and maybe mirror neurons are involved [12]. Of course, the effectiveness of MT in FNSD has to be evaluated empirically in future studies.

5 An integrative approach using functional and psychotherapeutic treatment modules seems promising. Though simple and one-sided conversion theories are outdated, emotional factors are important in FNSD. There has been a recent report on impaired emotion processing in FT [2], and a qualitative study showed life events as relevant for the majority of patients [19] even though it is not a sine qua non in DSM-5 anymore.

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Statement of Ethics

Written informed consent was obtained from the patient including video material.

Disclosure Statement

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Author Contributions

A.J. & R.H. treated the patient. A.J. wrote the draft of the manuscript. A.J. & R.H. discussed, wrote, and approved the final manuscript.

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