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Posttraumatic growth but not abnormal personality structure are typical for patients with essential blepharospasm

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ABSTRACT

Objective: The goal of our study was to find selective cognitive abnormalities in cognitive performance and personality profile of patients with essential blepharospasm (EB). Second, we wanted to see if in patients with EB we can identify posttraumatic growth (PTG) by comparison with a control sample and also with a sample of patients with hemifacial spasm (HFS).
Methods: We recruited 20 patients with EB, 20 patients with HFS and 23 demographically matched controls (NC). All participants (EB + HFS + NC) were assessed by the Posttraumatic Growth Inventory (PTGI) and a computer-based version of Cloninger's Temperament and Character Inventory (TCI-R). Furthermore, all participants underwent a brief battery of neuropsychological tests.
Results: EB patients had significantly higher scores in the PTGI questionnaire than those with HFS. As regards all TCI-R factors, there were no significant differences between EB, HFS or NC. In the cognitive battery, only in the Prague Stroop Test were patients with EB significantly impaired.
Conclusion: Our results are consistent with new information about a higher level of post-traumatic growth in EB patients than in HFS patients. We found no substantial evidence of a different personality profile in EB patients compared with HFS or NC.

1. Introduction

Dystonia is a syndrome consisting in sustained muscle contractions, frequently causing twisting and repetitive movements, or abnormal postures [1]. The pathophysiological changes which underlie dystonia include changes in the basal ganglia (BG) and their projections [2,3]. Dystonia can affect different muscle groups in the human body differentially (hence we refer to focal, segmental or generalized dystonia). Focal dystonia (FD) is characterized by contraction of one muscle or one muscle group. FD often appears on the face, neck or more acral parts of upper limbs. FD has a prevalence of 5/100 000 where essential (primary) blepharospasm (EB) [4] is one of its most frequent types. The age at onset of EB is usually in the 5th or 6th decade of life; with women slightly more affected than men. In EB the dystonic contraction impairs m. orbicularis oculi and causes sustained contractions of the muscles around the eyes (often shifting from one side to the other). The patients have normal vision, but for periods of time they are effectively blind leading to inability to drive, read or watch TV etc. EB is marked by unknown etiology and pathogenesis. However, it may come from impaired inhibition between BG and mesencephalic circuits, which results in functional damage to afferent and efferent parts of those circuits [2]. The most common treatment of EB consists in repeated injections of 4botulinum toxin (BTX) into dystonic muscles.

The status of primary FD as a purely motor disorder has recently been challenged as new evidence of its non-motor aspects has emerged. Together with this there is mounting evidence that the BG, beside motor control, play an important role in motivation and affective and cognitive functioning [5,6]. Some studies suggest that BG are essential also for the stability of personality traits [7]. It is therefore legitimate to surmise that, beside motor symptoms, patients with dystonia may suffer more often from psychiatric symptoms, that they may have abnormal personality traits or show

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cognitive changes [8]. As for the cognitive domain, some authors come forth with discordant results. The study by Jahanshahi, Rowe, & Fuller [9] stopped short of finding different cognitive performance between patients with focal dystonia and a control sample, whereas other studies report differences in executive functioning between FD and controls and between EB and controls [10–13]. A comprehensive study on cognition in EB found a broad cognitive involvement in measures of attention, visual search, visual-spatial abilities and memory (learning curve) together with changes in motor coordination tasks [14]. These alterations are explained as prefrontal and parietal dysfunction unrelated to the severity or duration of dystonia and independent of depression or anxiety [14].

EB may have a substantial impact on personality and mood [15,16]. Recent research has analyzed mainly negative effects: patients with FD had a higher rate of psychiatric comorbidities such as depression or obsessive-compulsive symptoms [17–20]. However, up to the present, there is not a single study of possible positive effects, especially of post-traumatic growth (PTG) in patients with EB. EB is so serious disorder that it can be seen as a repeated traumatic event with emotional sequelae which develop as a reaction to this traumatic experience. PTG may entail enhanced interpersonal relationships, new possibilities, increased personal strength, spiritual changes and greater appreciation of life [16]. A positive relationship has been identified between cognitive processing and PTG [21]. Most studies rate PTG with the Post-Traumatic Growth Inventory (PTGI; [22]), a 21-item questionnaire that assesses overall positive outcomes where its five factors distinguish between growth in the areas of new possibilities (factor 1), personal strength (factor 2), appreciation of life (factor 3), spiritual change (factor 4), and relating to others (factor 5). Patients with higher PTGI scores show consistently lower level of depressive and anxious symptoms in comparison to patients with lower PTGI score. [23].

A serious disease may be a trigger that contributes to a personal process of change and may serve as a stimulus, by which these changes can be promoted. Our theory of EB is based on the notion that EB initiates negative and positive changes in mood and personality of patients suffering from this serious disease.

Beside EB influence on mood these patients may have a specific personality profile mediated by BG system impairment, a factor that should also be taken into consideration [24]. Those patients' results show consistent changes in the personality profile of patients with FD; however, a more specific study of the personality profile of patients with EB is so far unavailable. Lencer et al. used for this purpose the NEO FFI [25], a personality inventory measuring global personality dimensions, such as Neuroticism, Extroversy and Openness. However, the assumption that the NEO FFI may be associated directly with BG dysfunction has so far not been proved by previous research. The literature on this topic lacks studies that would use instruments with proven association between personality traits and BG dysfunction, as in the case of TCI-R [7].

Hemifacial spasm (HFS) is a unilateral, involuntary, irregular clonic or tonic movement of muscles innervated by the 7th cranial nerve. It is most often caused by vascular loop compression at the root exit zone of the facial nerve. HFS is not classified as dystonia, it is considered to be a bundle of intermittent, involuntary and synchronic myoclonic twitches, whose origin is so far not consistently explained and is presumably caused by peripheral mechanisms. HFS's onset is frequently in the 4th or 5th decade of life with no gender differences [26,27]. HFS does not impair visual perception, however, like EB, it may have psychosocial consequences. In contrast to EB we do not suppose HFS to be related to BG systems or to other areas such as are essential in cognitive abilities or neuropsychiatric disorders [28]. However, HFS patients undergo the same BTX treatment as EB patients.

Hence, the goal of our study was to find selective cognitive abnormalities in cognitive performance and personality profile, especially in the temperament of patients with EB. Second, we wanted to find out if such changes are associated with BG dysfunction. Third, we wanted to see if in patients with EB we could identify PTG comparing with control sample also with a sample of HFS patients.

2. Methods

2.1. Subjects

Our study used twenty patients with EB, twenty patients with HFS and twenty-three demographically matched controls (NC). All subjects were recruited from the Movement Disorders Center, Department of Neurology, First Faculty of Medicine and General University Hospital in Prague. All patients were examined by a neurologist specialized in movement disorders. Both clinical samples (EB + HFS) were treated with and underwent neuropsychological assessment between the fourth to sixth weeks after BTX, when the most effective treatment effect could be expected. In addition, all patients completed a self-rating scale on the efficacy of BTX treatment (excellent, good, satisfactory, very poor) and on the influence of EB on the activities of daily living (ADL) [29] in patients with EB only (Jankovic Rating Scale (JRS) and Blepharospasm Disability Index (BSDI)) [30] in both (EB + HFS). JRS is a domain-specific rating scale for the evaluation EB severity, BSDI is instrumental activities of daily living rating scale specialized on the evaluation of driving a vehicle, reading, watching TV, shopping, walking, doing everyday activities. The NC sample was demographically matched based on non-random sampling. The subjects were employees of the First Faculty of Medicine and General University Hospital in Prague. The study was approved by the General University Hospital IRB, and all participants gave their written informed consent.

2.2. Inclusion criteria

These were as follows: absence of any comorbidity in EB and HFS patients based on anamnestic evaluation and neurological examination, absence of dementia according to the MMSE, absence of signs of atypical or secondary parkinsonism, severe or unstable depression, anticholinergic medication, absence of other medical or neurological conditions potentially causing cognitive impairment (e.g., seizure, stroke, or head trauma).

2.3. Neuropsychological assessment

All participants (EB + HFS + NC) were assessed with the Post-traumatic Growth Inventory (PTGI, [16]) and with a computer-based version of Cloninger's Temperament and Character Inventory (TCI-R) [31]. The PTGI is self-rating inventory in which higher scores indicate higher posttraumatic growth in several domains. All participants also underwent a brief neuropsychological battery, which included Mini-Mental State Examination (MMSE) [32], test of phonemic verbal fluency [33], Prague Stroop Test [34] and Frontal Assessment Battery (FAB) [35]. For the evaluation of neuropsychiatric symptoms we used Spielberger's State-Trait Anxiety Inventory (STAI X1/X2) [36] and Beck's Depression Inventory, Second Edition (BDI-II) [37].