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Psychiatric Symptoms Associated with Focal Hand Dystonia

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Abstract

Myoclonus dystonia and idiopathic dystonia are associated with a greater frequency of obsessive compulsive disorder (OCD) and major depression. We investigated the frequency of OCD in 39 patients with primary focal hand dystonia (FHD) using a semistructured interview. OCD and subsyndromal OCD was diagnosed in 5 of 39 (12.82%) patients with FHD, whereas OCD occurs in 2.3% of the general population. Recurrent depression occurred in (7 of 39) 17.95% of patients with FHD along with a family history of depression in (16 of 39) 41.02%. Overlapping mechanisms manifesting as FHD may also predispose to OC symptoms and likely implicates a common striatal dysfunction.

Keywords

obsessive compulsive disorder; depression; dystonia; striatum; obsession; compulsion

Focal hand dystonia (FHD) is characterized by taskspecific sustained muscle contraction that leads to impaired use. Movement disorders such as Parkinson's disease, Tourette's syndrome, or Huntington's chorea are commonly associated with comorbid neuropsychiatric symptoms related to the underlying neurobiology of the movement disorder. More severe

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forms of dystonia have been associated with obsessive compulsive disorder (OCD). For instance, myoclonus dystonia and DYT 11^{1,2} have been associated with a greater frequency of OCD. OCD had also been reported to be more frequent in idiopathic dystonia patients and their family members,³ although no association with DYT1⁴ has been observed. Blepharospasm has also been associated with a greater number of obsessive compulsive symptoms.⁵ In a study of 86 patients with primary cervical dystonia or blepharospasm, the odds ratio of having OCD was 8.4 compared with their control group.⁶

OCD is a neuropsychiatric disorder characterized by obsessions or compulsions reflecting frontostriatal dysfunction. Obsessions are recurrent uncontrollable thoughts or impulses associated with anxiety or distress. Compulsions are repetitive behaviors or mental acts that are performed in response to an obsession or set of rules and aimed to reduce distress. For the diagnosis of the disorder of OCD, the symptoms must be associated with marked distress, occur more than 1 hour per day, or significantly interfere with social or occupational functioning.⁷

FHD has been associated with both striatal and cortical pathology with mechanisms of decreased inhibition, greater plasticity, and sensory impairments.⁸ Pathology affecting the basal ganglia includes disorganized putaminal somatotopy,⁹ larger putaminal volume,¹⁰ and hyperechogenicity in the lenticular nucleus.¹¹ OCD is also characterized by frontostriatal dysfunction particularly implicating structural and functional abnormalities of the orbitofrontal-striatal circuitry along with abnormalities in tasks associated with striatal function such as implicit sequence learning, procedural learning, and response reversal.¹² In this study, we investigated the relationship between OCD and FHD by assessing the frequency of subsyndromal and syndromal OCD in patients with FHD.

PATIENTS AND METHODS

Patients with FHD were recruited from a convenience sample of patients who had been seen at the Human Motor Control Section (HMCS) clinic or at the Botox clinic at the National Institute of Neurological Disorders and Stroke, National Institutes of Health (NIH). Over the 6-month period of the study, subjects who were being seen at the Botox clinic were contacted prior to their appointment for either a same day assessment or to make a new appointment. Newly diagnosed subjects on the HMCS database (diagnosed within the last 2 years) were also contacted. Inclusion criteria included patients >19 years of age with FHD and no other serious neurological or medical illnesses. The study was approved by the NIH Institutional Review Board and patients signed informed consent.

Subjects underwent a semistructured interview by a psychiatrist or psychologist for the assessment of psychiatric disorders (Structured Clinical Interview for the diagnosis of DSM-IV Axis I disorders). Subsyndromal OCD was identified if patients fulfilled all criteria for OCD except for the criterion of functional impairment. All identified cases were reviewed with an expert in OCD for diagnostic confirmation. Patients were assessed with the clinician-rated Yale-Brown Obsessive Compulsive Scale (Y-BOCS), a 10-item scale in which each item is rated from 0 to 40.¹³ The scale assessed the amount of time spent and the impairment, distress and degree of resistance, and control over the obsessions and

compulsions. A score of 0 to 7 is subclinical, 8 to 15 mild, 16 to 23 moderate, 24 to 31 severe, and 32 to 40 extreme. Family psychiatric history was questioned based on any known diagnoses of depression, alcohol abuse, bipolar disorder, or schizophrenia that interfered with function in siblings, parents, or grandparents but the diagnosis was not confirmed with diagnostic criteria or assessment of the family member. Subjects also completed the Beck Depression Inventory, the Beck Anxiety Inventory, and the Sheehan Disability Scale.

RESULTS

Forty patients with FHD were assessed [mean age 48.1 (SD 10.3), 14 women, FHD duration mean 12.14 (SD 9.12) years] from 68 patients contacted. Twenty-eight patients did not enter the study because of lack of interest, lack of time, or inability to schedule an appointment that suited their schedule as the majority of patients were working. Twenty-seven patients had dystonia secondary to writing, 12 patients had dystonia secondary to use of a musical instrument, and 1 patient had dystonia secondary to typing. Thirty-nine of forty patients had primary dystonia and 1 patient had dystonia secondary to head trauma with secondary FHD and focal epilepsy. This patient's family history was positive for epilepsy but negative for dystonia. Ten of forty patients had a family history (including cousins) positive for either tremor or dystonia. At the time the study was conducted, there was no indication for genetic testing. Six of forty patients were on an antidepressant for depression, anxiety, or pain and 6 of 40 were on an anticonvulsant for pain with one for a seizure disorder. Twenty of forty were receiving regular Botox treatment for their FHD symptoms.

In the following, we base the analysis on patients with primary FHD (N = 39). Lifetime OCD was identified in 4 of 39 (10.25%) with current OCD in 3 of 39 and subsyndromal OCD in 1 of 39 (2.56%) with symptoms preceding the dystonia onset. Six of thirty-nine (15.38%) patients had generalized anxiety disorder; 2 of 39 (5.13%) had simple phobia and 3 of 39 (7.69%) had social phobia with fear of public speaking that preceded the onset of dystonia; and 1 of 39 (2.56%) had panic disorder. The diagnoses of anxiety disorders overlapped with a total diagnosis in 10 of 39 (25.64%). A lifetime history of major depression was identified in 10 of 39 (25.60%) with recurrent major depression in 7 of 39 (17.95%), current major depression in 3 of 39 (7.69%), and current dysthymia in 2 of 39 (5.13%). In total, 13 of 39 (33.33%) patients had a previous or current history of a depressive disorder. All patients with OCD and subsyndromal OCD had comorbid depression. One patient had a diagnosis of a substance use disorder. There were no diagnoses of psychotic disorders, bipolar affective disorder, or post-traumatic stress disorder. There was a family history of depression in 16 of 39 (41.02%), "nervous breakdowns" in an additional 3 of 39 (7.69%), and alcohol use disorders 8 of 39 (20.51%). In the following, we only assess differences between OCD and non-OCD in primary FHD and do not include subsyndromal OCD. As expected, the Y-BOCS total score was greater in patients with OCD [17.25 (SD 5.40)] than without [0.96 (SD 2.4)] (t = 7.95, df = 36, P < 1000.0001). BAI scores were also higher in the patients with OCD [11.25 (SD 5.90)] compared with those without [4.75 (SD 5.01)] (t = 2.41, df = 36, P = 0.02). There was a trend toward higher BDI scores in patients with OCD [11.25 (SD 6.70)] compared with those without [5.75 (SD 5.43)] (t = 1.87, df = 36, P = 0.07). There were no differences in Sheehan

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Disability Scale scores in patients with OCD [8.21 (SD 4.29)] compared with those without [7.10 (SD 7.01)] (t = 0.31, df = 36, P = 0.77).

DISCUSSION

In this study, we demonstrate a frequency of OCD of 10.25% and subsyndromal OCD of 2.56% in patients with primary FHD. This frequency contrasts with the lifetime prevalence of 2.3% of OCD in the general population.¹⁴ Our study results dovetail with previous reports of elevated frequency of OCD in patients with more severe forms of dystonia such as idiopathic dystonia and myoclonus dystonia.^{1,3,6} The lifetime prevalence of major depression in primary FHD was 25.60%, recurrent major depression was 17.95%, and a family history of depression in immediate relatives was 41.02% in FHD. These rates contrast with the lifetime prevalence of major depression in the general population of 17%.¹⁵ Recurrent major depression is associated with DYT1 with a similar rate of expression in manifesting carriers (13.5%) as nonmanifesting carriers (13.3%) when compared with noncarriers (4.6%) (odds ratio of development of OCD in carriers versus noncarriers = 3.04). Although we did not intend to focus on other psychiatric disorders, recurrent major depression along with a family history of depression may be similarly elevated consistent with other reported studies of an association between idiopathic dystonia and DYT1 with recurrent major depression.¹⁶ We caution that this is a small sample size without a control group and more extensive studies with a larger population along with more systematic assessment of family members would be necessary. Our study suggests overlaps in the pathophysiology of OC symptoms and FHD likely implicating a common striatal dysfunction. The pathophysiology may be related to a similar underlying genetic diathesis or the neurobiology of FHD may affect similar striatal regions to that of OCD. These observations are convergent with the greater comorbidity of psychiatric disorders observed in patients with other movement disorders.

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Author Roles: Valerie Voon: research project: conception, organization, and execution; statistical analysis: design, execution, and review; manuscript: writing and review. Tracy Butler: research project: execution; manuscript: review and critique. Vindhya Ekanayake: research project: execution; manuscript: review and critique. Cecile Gallea: research project: execution; manuscript: review and critique. Cecile Gallea: research project: execution; manuscript: review and critique. Dennis Murphy: research project: execution; manuscript: review and critique. Mark Hallett: research project: conception; statistical analysis: review; manuscript: review.

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