

IS PSYCHOGENIC DYSTONIA A VALID DIAGNOSIS?

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Introduction: By convention, since roughly 1950-1975, dystonia has been considered a neurological disorder whose cause does not require any psychological abnormality. However, a small percentage of patients with dystonic-like movements have unusual features that suggest their abnormal movements may be a medically unexplained pseudoneurological behavior. Early researchers described a range of dystonia presentations with typical "organic" features at one end of a spectrum and inconsistent, unusual, neurotic or dramatic patients at the other end. In 1975 Fahn and Eldridge stated that there had been no proven case of "psychogenic dystonia," meaning a case that looked like dystonia but either was clearly malingering or a case in which symptoms were cured by psychotherapy, suggestion or placebo. Over the next decade they found and described several patients who were the exception to this rule. Unintentionally this approach created a new diagnosis, psychogenic dystonia, with a case definition based mostly on treatment response. The obvious question is, is this new diagnosis any good? In this presentation I will review what we know about whether "psychogenic dystonia" is a useful diagnosis, and present some relevant retrospective data from our center.

Methods: *Methods: review.* Robins and Guze (1970) detailed a method for establishing diagnostic validity in five phases. I gathered references pertinent to these five phases. The search was nonsystematic but included MEDLINE searches, book chapters, and discussions with colleagues. *Methods: retrospective study.* We retrospectively identified 58 patients judged by their treating neurologist to have atypical ("psychogenic") dystonia and 105 control patients with typical dystonia out of 1565 dystonia patients in our electronic database. Charts from both atypical and control patients were examined for features selected from prior literature on psychogenic dystonia or hysteria, and from our experience. Chi-squared tests were performed to determine significance. Survival analyses were completed for both groups based on site of onset, and hazard ratios compared relative risk of spread across groups.

Results: *Results: review.* There are very limited data to address validity in terms of laboratory studies, delimitation from other disorders (most especially from typical dystonia and established psychiatric diagnoses). Only a couple of reports address follow-up, treatment response and family history.

Results: retrospective study. Prevalence of many of the identified variables differed significantly in the atypical dystonia group. These variables can be sorted into a few categories: (1) routine history or exam features that could be as easily interpreted without the "psychogenic" label; (2) features suggesting malingering or somatization disorder; (3) suggestibility, distractibility, excessive sensitivity, excess pain, presence of non-anatomical neurological dysfunctions, and increased symptoms with hyperventilation; (4) certain criteria suggesting other psychiatric illnesses. Family history of dystonia does not differ significantly between groups. However, some of these features are unlikely to be clinically useful. The presence of other psychiatric illness or an unstable family, for instance, showed a low positive predictive value (PPV). Other features, such as bizarre gait, had a higher PPV. Bodily distribution of abnormal movements was similar between the groups at presentation, but in atypical patients the abnormal movements were significantly more likely to spread to involve other body parts.

Discussion: Fahn and colleagues have done a great service by drawing attention to these patients and by identifying features important for further clinical study. However, issues of diagnosis matter greatly to patient care. Our clinical study has limitations, including primarily its retrospective nature and the assignment of patients to diagnostic groups by clinical judgment alone rather than by reference to any operationalized diagnostic criteria. Both these limitations may bias ascertainment. Nevertheless, available data suggest caution in how confident one can be in clinical diagnosis of psychogenic dystonia. The diagnosis of psychogenic dystonia is problematic for other reasons. The word "psychogenic" adds no real meaning. Although Fahn and colleagues defined psychogenic dystonia by spontaneous remission or response to placebo or psychotherapy, these are seen in a minority of typical dystonia patients. The presence of concomitant psychological symptoms has low PPV. The assumption that atypical illnesses are psychologically caused can short-circuit the need for continued clinical observation in the patients who need it most. Finally, misdiagnosing psychogenic dystonia can harm the patient.

I will discuss an alternative though not original proposal that avoids presumption of etiology but accepts that many of these patients probably have a different illness from typical ("real") dystonia. At this time there are no data to support a claim of superior outcome, yet since there are few outcome data at all, it seems reasonable to consider alternatives.

I recommend limiting diagnosis to deciding whether the abnormal movements are either (a) not dystonia, (b) possible dystonia but with unusual features, e.g. phenomenologically atypical, placebo-responsive, or exacerbated by emotional stress; or (c) so close to dystonia phenomenologically as to be indistinguishable based on the abnormal movements alone. We can add diagnostic information for patients with validated diagnoses such as DSM-IV somatization disorder or major depression.

Randomized controlled trials will be required to test specific treatments. The report by Voon and Lang (2005) suggests one pharmacological approach for some patients. As far as psychological management, an atheoretical and rehabilitative approach sidesteps many of the issues that render patients hesitant to accept a diagnosis of psychogenic dystonia or imputation of their symptoms to stress. I will discuss how we approach these patients at the Washington University Movement Disorders Center.

This approach can fit with an evidence-based or at least a modest, atheoretical approach to medical education. This approach also suggests a research agenda for diagnosis, pathophysiology, and treatment. It may or may not prove to be ideal, but it offers a number of testable hypotheses.

In conclusion, a small but real fraction of patients with abnormal movements resembling dystonia have features suggestive of somatoform illness. However, there is little evidence to support the diagnostic validity of "psychogenic dystonia" as currently defined. Further research is sorely needed.

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