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Increased variability in spiral drawing in patients with functional (psychogenic) tremor

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ABSTRACT

Increased variability is a characteristic clinical and physiologic feature of functional (psychogenic) tremor. In this study, we use computerized spiral analysis to show that the variability of a motor task is a quantifiable characteristic of functional tremor. We compare functional tremor patients to phenomenologically similar dystonic tremor patients and to normal controls. We used the spiral severity score, a measure that does not incorporate spiral tightness, as a marker of spiral drawing performance, and inter-spiral tightness variability (based on the 25–75%^{ile} range in tightness across ten spirals) to evaluate the effects of functional tremor on drawing spirals. The spirals of 74 participants: 22 functional tremor, 21 dystonic tremor, and 31 normal controls were analyzed. Spiral severity was higher in both tremor groups compared to controls, but did not differentiate them. Inter-spiral variability, however, was higher in the functional tremor group compared to both other groups. Thus, spiral analysis captures variability of a motor task and may be used as an objective test for functional tremor. The effect of functional tremor in other motor tasks should be investigated.

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1. Introduction

Psychogenic or functional movement disorders (FMD) can be defined as those movements thought to arise from psychological factors rather than having an organic etiology (Fahn, Jankovic, & Hallett, 2011). They constitute a significant diagnostic and treatment dilemma for the clinician (Hallett, 2006), and their pathophysiology is poorly understood and controversial (Stone & Edwards, 2011). Increased variability and inconsistency of movements are characteristic clinical features of FMD, and are elements of the required criteria for the diagnosis of clinically established or probable FMD (Fahn et al., 2011).

Tremor is defined as rhythmic oscillations of a part of the body, usually around one or more joints (Hess & Pullman, 2012). There are many types of tremor disorders characterized (but not always distinguishable) by frequency, regularity, directionality and the tasks or states during which they occur (Deuschl, Raethjen, Lindemann, & Krack, 2001). Essential tremor is the most common, a slowly progressive monosymptomatic disorder characterized by moderately high frequency 8–10 Hz kinetic and postural tremors, predominantly involving the hands, that decrease in frequency with age (Brennan, Jurewicz, Ford, Pullman, & Louis, 2002). Other types of organic tremors include 4–6 Hz rest tremor in Parkinson disease, 2–6 Hz cerebellar intention tremor, 4–8 Hz dystonic tremor, 8–12 Hz enhanced physiologic tremor, and 14–18 Hz orthostatic tremor (Deuschl et al., 2001). Functional tremor is the most common type of FMD (Fahn et al., 2011), and its diagnosis is amenable to confirmatory physiologic testing (Piboolnurak et al., 2005; Schwingenschuh et al., 2011). Findings such as increased tremor amplitude with inertial weighting, distractibility, and entrainment with other rhythmic movements can help to confirm the diagnosis in FMD in suspected cases. Importantly, there is also an increased variability in the basic characteristics of tremor that is notable over the short period of time during testing (Schwingenschuh et al., 2011). This increased variability is helpful in distinguishing functional from organic causes of tremor such as dystonic tremor that phenomenologically may appear similar. In dystonic tremor, the oscillations may be irregular in amplitude and occur at a range of frequencies (Deuschl et al., 2001) but the irregularities do not vary greatly in a given patient during evaluation. Dystonic tremor is thought to occur secondary to the sustained co-contraction of agonist and antagonist muscles and/or compensatory muscle contractions, and are consistently replicated by specific tasks such as writing, e.g. writer's cramp (Fahn et al., 2011).

Spiral drawing has been utilized for years as a clinical tool to observe tremors and other abnormal movements. Spirals can be analyzed by visual inspection, or by computerized analysis, a method of quantifying upper limb motor function from handwritten spirals using a digitizing tablet and writing pen connected to a computer (Pullman, 1998). This method records the pen x and y positions, force and time data, without wires or other attachments. It is easily performed over several trials. Recently, computerized spiral analysis with a digitizing graphics tablet has been used to evaluate many different types of neurological disorders (see review (Van Gemmert & Teulings, 2006)) including multiple sclerosis (Longstaff & Heath, 2006), dyskinesia (Liu, Carroll, Wang, Zajicek, & Bain, 2005), Parkinson's disease (Saunders-Pullman et al., 2008), essential tremor (Louis et al., 2012) and Niemann-Pick Disease (Hsu et al., 2009).

Quantification of handwritten spirals is based on "unraveling" the spiral (Fig. 1) through a radius-angle transformation from which kinematic, dynamic and physiologic features are derived mathematically. After the spiral (x, y) data points are acquired, the radius-angle transformation is defined as $x = r \sin \theta + x_0$, $y = r \cos \theta + y_0$; so that the radius r is as follows: $r = \sqrt{(x - x_0)^2 + (y - y_0)^2}$, arc tan $\theta = (y - y_0)/(x - x_0)$. In these equations (x_0, y_0) is the spiral center, (x, y) is the spiral coordinate in image plane domain and (r, θ) is its counterpart in the polar expression. This simplifies to the polynomial relationship: $\theta = a\ r$, where a is a positive constant. The spiral center is determined by taking the first non-zero pen pressure measurement and adjusted using an optimization procedure to reduce low frequency artifact (Wang et al., 2008).

An array of spiral indices are computed from the radius-angle transformation that relate to clinical phenomena, e.g. drawing smoothness, drawing speed, acceleration, decrement, tremor, spiral tightness, and ataxia (Hsu et al., 2009). Spiral tightness correlates with micrographia or macrographia, and is a measure of the number of spiral loops normalized per cm. An overall spiral severity score,