Movement Disorders
CLINICAL PRACTICE

CASE SERIES

Positional Tremor and its Treatment

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Abstract: Background: Positional tremors arise when a patient's tremor is brought on during specific positioning of the involved body part. They can be distinguished from postural tremor, wherein a patient's tremor is elicited in any posture, and from task-specific tremor, wherein a patient's tremor occurs only during a certain task. **Cases:** We describe 2 cases of positional tremor that were markedly improved with botulinum toxin injection. **Discussion:** The term "positional" is a valuable descriptor for tremors. In patients with positional tremor, botulinum toxin may be beneficial for treatment. Lidocaine injection provides a transient way to test for the appropriateness of botulinum toxin injection in these patients.

Tremor can be characterized in terms of topographic distribution, frequency, associated anatomy or disease process, or activation condition.[1] Activation condition is classically broken down into those tremors arising during rest or action (posture, intention, or kinetic movement), but these distinctions oversimplify an often complex pattern. One type of "activation condition" that has been largely lumped in with others is position. Positional tremors arise when a tremor is brought on during specific positioning of the involved body part. They can be distinguished from postural tremor, wherein a tremor is elicited in any posture, and from task-specific tremor, wherein a tremor occurs only during a certain task. To complicate matters, postural tremors may be present more in one position than in another.[2] Likewise, positional tremors may present themselves during specific tasks, such as putting on makeup or pitching a baseball; however, on deeper review, it becomes clear that the patient can induce the tremor by putting their body part in the associated position without actually performing the task. Task-specific tremors are defined as occurring *primarily* during a specific task and may be present to a lesser degree during other uses of the hands.[1] Positional tremors, by contrast, are present with equal intensity during any task that involves a certain position. Thus, there is a complex fluidity to the terminology of tremor. Here, we discuss two patients with positional tremor. We argue that a nuanced history and examination can distinguish this entity from other tremor types and propose that botulinum toxin injections should be considered for position-dependent tremors, regardless of the underlying etiology.

Case Series

Case 1

A 50 year-old, right-handed woman presented to the Yale Movement Disorders Clinic reporting right arm tremor. She described the tremor as mostly interfering with her ability to put on makeup. It was not present when she was performing tasks with her hand away from her body, such as writing or using utensils. Her medical history was significant for cervical dystonia and associated dystonic tremor in her head, for which she received botulinum toxin injections. She was on clonazepam 0.125 mg twice daily for her dystonia, which helped with her head tremor, but not with her arm tremor. She had a family history of Parkinson's disease in her maternal grandfather and head/hand tremor and cervical dystonia in multiple family members. On examination, she exhibited a proximal right arm tremor that was present when she brought her hand close to her face, with shoulder adducted and arm flexed (Video S1). This occurred irrespective of the task (putting on makeup, drinking from a bottle, placing hand over face). It was not present in any other position. When the shoulder was stabilized, the tremor subsided. When the patient's arm was brought to the position passively (i.e., with intentional component removed) and then released while the

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patient maintained that position, the tremor still occurred. There was no dystonic posturing in her hands or arms. There was no vocal tremor or left arm tremor. She exhibited cervical dystonia characterized by left laterocollis and "no-no" head tremor when her head was held in the neutral position (straight ahead). The head tremor was exacerbated when she turned her head to the right. The remainder of her neurologic examination was unremarkable. With electromyography (EMG) guidance, we identified the motor endplate zone in the right biceps muscle by seeing motor unit potentials with initial negative phase, and 2 mL of 1% lidocaine was injected at the site. Her examination 30 minutes after injection demonstrated improvement in her tremor that persisted for several hours. Given this success, she underwent injection with 50 units of onabotulinumtoxinA into the right biceps. Two weeks after the injection, her tremor was markedly improved.

Case 2

A 20 year-old, right-handed man presented to the Human Motor Control Section at the National Institute of Neurological Disorders and Stroke for right middle finger tremor. He was a high-performing baseball pitcher with an intensive training history for 8 years. Four months before his presentation, he had noticed twitching in his right middle finger while pitching, when applying force to the ball with his middle and ring fingers toward the end of a pitch. He lost control of the ball, and it veered off to the left. His forearm also felt very fatigued early in the practice, which was uncharacteristic for him. He later noticed that, when flexing the middle finger, the tremor would emerge, even when he was not pitching. The twitching would also occur after lifting weights. His grip was unaffected. The problem persisted even after a 2-week break from pitching. EMG and nerve conduction studies did not show evidence of median, radial, or ulnar neuropathy. He had no pertinent medical history. His family history was significant for Parkinson's disease in his paternal grandfather. His examination demonstrated tremor in the right middle finger during flexion of the metacarpophalangeal and proximal interphalangeal joints, as well as a fine postural tremor in his right hand when arms and hands were extended. Flexor digitorum superficialis activity was identified by the isolated flexion movements of the right middle finger. The motor endplate zone of the third finger fascicle was localized under EMG guidance and injected with 1 mL lidocaine 0.5% solution. The baseline video could not be retrieved. The video demonstrates improvement in tremor after the lidocaine injection, and subsequent resolution of tremor with injection of botulinum toxin (Video S2).

Discussion

The term "positional" is a valuable descriptor for tremors. Some tremors previously described in the literature as task-specific might have been better described as positional. For example, Rothwell et al. (1979) labeled one patient's tremor as a primary writing tremor, but the tremor was present during any activity that involved wrist pronation, not just with writing.[3]

The word "positional" is a descriptor and does not imply tremor etiology. In this series, the first patient (Case 1) had dystonia of an adjacent body part but did not exhibit dystonic posturing of the tremor-affected limb. Nevertheless, dystonic etiology underlying tremor could not be ruled out. The second patient (Case 2) had an associated task that he had performed repetitively and with high intensity (pitching), but the tremor always occurred in the flexed position of the affected finger regardless of the task (similar to the patient described by Rothwell and coworkers), and there was no evidence of dystonia. In both cases, the tremor was first noted by the patient during certain tasks, but examination demonstrated that it was the specific position of the affected body part, not the task, that was the driving force. Whether task association in positional tremors represents an evolution from pure task-specific tremors to positional tremors or whether positional tremors may become more task-specific over time remains to be determined.

Several studies have explored botulinum toxin injections in patients who have position-dependent tremor, regardless of the underlying etiology (Table 1). Saifee et al. conducted a pilot study assessing botulinum toxin injections in 8 patients who had position-dependent upper limb action tremors.[4] Six of those 8 patients (75%) reported that they were "improved" or "much improved." Similarly, Kim et al. retrospectively reviewed the

TABLE 1 Overview of published studies assessing botulinum toxin for the treatment of position-dependent tremor

Study	Type of study	No. of patients	Tremor location	Position that induces tremor	Neurologic comorbidities	Outcome measures	Response to treatment
Saifee et al., 2016[4]	Prospective, uncontrolled	8	Upper limb	Supination or pronation with elbow flexed	Dystonia (1 patient), head tremor (2 patients)	CGI, functional change	75%, Improved or much improved; 25%, no change; 62.5%, ADL improvement
Kim et al., 2014[5]	Retrospective analysis	19	Proximal upper limb (shoulder or elbow)	Targeting nose	Varied: Dystonia, ET, TBI, stroke, MS, PD	Observable tremor improvement, functional change	63%, Moderate or marked improvement; 26%, mild; 11%, none

CGI, clinical global impression; AD, activities of daily living; ET, essential tremor; TBI, traumatic brain injury; MS, multiple sclerosis; PD, Parkinson's disease.

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efficacy of botulinum toxin injections in 19 patients with position-dependent upper limb tremors.[5] Twelve of those 19 patients (or 8 of 11 patients without dystonia) demonstrated moderate or marked improvement in the tremor, based on a combination of phenomenological and functional response. Both studies used EMG guidance and examination to tailor the injection pattern to the individual patient.

Although the etiology may differ between patients with positional tremor, we postulate that abnormal muscle spindle afferent drive may be the common pathophysiological thread in these cases. This hypothesis is further supported by the treatment response in our patients. Lidocaine reduces the activity of intrafusal (spindle) γ motor fibers and relaxes the spindle.[6] This relaxation decreases the afferent drive from the spindle to the spinal cord, thereby preventing tremor activity. Lidocaine blocks γ motor fibers at lower concentrations than extrafusal α motor neurons.[6] Thus, lidocaine can relax the spindle without causing muscle weakness. Botulinum toxin exerts its effect similarly by blocking the γ motor junctions at the motor endplate without causing a major block of the extrafusal fiber junctions.[7] Therefore, we administered lidocaine as a test and justification for botulinum toxin use. Indeed, our patients experienced improvement in tremor without weakness after lidocaine and, subsequently, after botulinum toxin injection.

In individuals with position-dependent tremors, etiology may be less pertinent to the treatment strategy than the phenomenology and categorization as position-dependent. Botulinum toxin injection has been shown to be efficacious in positional upper limb tremors in several previous studies, and the findings in our patients bolster those results. These tremors may be particularly amenable to injection treatments due to the relative ease with which a tremulous "driver," or responsible muscle, may be identified; in both of our patients, the tremor was markedly improved with injection into only one muscle. Lidocaine injection proved to be a valuable and more transient trial procedure before botulinum toxin injection. In clinical practice, lidocaine injection may allow practitioners to test whether they will influence the tremor without undue weakness before initiating botulinum toxin injections. Further exploration into position-dependent tremors as a distinct entity will assist the neurologic community in identifying patients for whom botulinum toxin injection may be particularly beneficial.

Author Roles

1. Research Project: A. Conception, B. Organization, C. Execution; 2. Statistical analysis: A. Design, B. Execution, C. Review and Critique; 3. Manuscript preparation: A. Writing the First Draft, B. Review and Critique.

S.M.S.: 1A, 1B, 1C, 3A, 3B
M.H.: 1A, 1B, 1C, 3B
1B, 1C, 3B
D.B.D.: 1C, 3B
S.T.: 1A, 1B, 1C, 3B

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Supporting Information

Videos accompanying this article are available in the supporting information here.

Video S1: Case 1. This video shows the patient in the following scenarios: 1) baseline, at her initial visit and just before lidocaine injection; 2) 30 minutes after lidocaine injection; and 3) 2 weeks after botulinum toxin injection.

Video S2: Case 2. This video shows the patient in the following scenarios: 1) after lidocaine injection and 2) after botulinum toxin injection.

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