

Primary Task-specific Bowing Tremor: An Entity of its Own?

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A professional violinist in his early 60s, playing in a prestigious German orchestra for more than 20 years, presented to our institute because of a task-induced tremor in his right arm when playing the violin. We describe the phenomenology of this tremor and its treatment options and compare it to findings in primary writing tremor (PWT). We then discuss whether primary bowing tremor is an entity of its own (similar to PWT) and propose hypotheses that would derive from such a definition. *Med Probl Perform Art* 2012; 27(4):224-226.

CASE PRESENTATION

A male violinist in his early 60s had been playing the violin for over 50 years, with more than 20 years in a professional German orchestra. Tremor appeared first at the age of 46, in the right arm, while only playing the violin. Prior to this he was involved in a car accident, resulting in a late arrival for a concert where he had to play numerous, long-lasting, fast, and repetitive bowing-movements (16th notes) in a long-lasting, romantic symphony. The following day he developed a pain syndrome in the right shoulder. Subsequently, he recognized a progressive involuntary tremor of the right arm while playing the violin. From 2005 onward, he has received medical treatment for continued pain syndromes in the musculoskeletal system.

In 2010, he presented to our institute for the first time because of accelerated tremor progression for 1 year, which had caused him to reduce or cancel solo- or chamber music performances.

Clinical neurological examination did not reveal any pathologic findings. Reflexes were symmetrical without pathologic reflexes. No sensory deficit was detectable. Pal- laesthesia was normal. Diadochokinesis was undisturbed. There were no signs of cerebellar dysfunctions and no rigor

or bradykinesia present. Cranial nerves were normal. No resting tremor was visible on either side. Smelling disorders were not present.

Further diagnostic measures including a dopamine transporter (DAT) scan and a cerebral and cervical-spine MRI were unremarkable. Electrophysiological measures were normal for motor evoked potentials (MEP) and marginally prolonged for somatosensory evoked potentials (SEP).

In the family history, his father is reported to suffer from either writer's cramp (WC) or primary writing tremor (PWT).

When playing the violin, a pronation-supination tremor as well as a flexion-extension tremor in the wrist of about 7-8 Hz was visible and audible (Fig. 1), especially when playing slow legato notes. Tremor was visible on all four strings of the violin equally and was most present between the middle and nut of the bow. No postural or resting tremor was visible. When the left hand was used for bowing, no tremor appeared.

Tremor appeared in everyday situations (e.g., filling a glass of water) about 1.5 years ago, however to a lesser extent and unilateral as well.

Tremor initially responded to beta-blockers (propranolol 20 mg); however, this effect wore off over the course of 1 year. Alcohol still improves symptoms. Trihexiphenidyl treatment did not lead to an improvement. Injections of botulinum toxin (Dysport®) were given twice and had a positive effect in everyday life activities and some effect when playing the violin, without side effects. The first injections were given to the bicep, tricep, pronator teres, and supinator muscles. The second injections were localized to the radial extensor carpi, radial flexor carpi, supinator, bicep, and tricep muscles. Treatment was not continued, because improvement at the violin was insufficient.

Primidone (anticonvulsant) led to an improvement, but due to side effects (tiredness, slowing, and erectile dysfunction), continuous treatment had to be stopped. He currently takes primidone 75 mg prior to playing solos or chamber music. Neither gabapentin nor topiramate (anticonvulsants) was effective. Clozapine was proposed, but so far not taken. Deep brain stimulation was declined by the patient.

The patient continues to play in the orchestra and has again started to perform chamber music concerts, however less frequently than before tremor onset.

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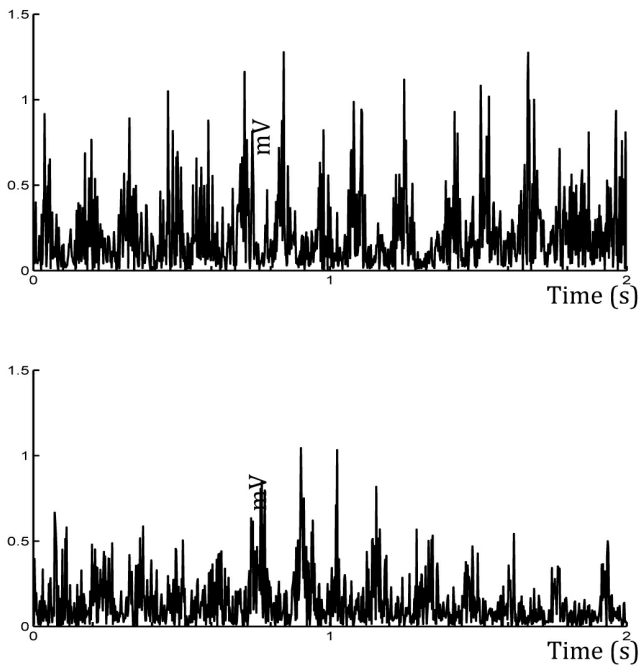


FIGURE 1. Rectified EMG of the flexor carpi radialis (*upper panel*) and the extensor carpi radialis (*lower panel*) with a reciprocal activity of about 7–8 Hz.

DISCUSSION

Task-Specific Tremor

Tremor is defined as a rhythmical, oscillatory, and involuntary movement of a body part¹ and is one of the most common movement disorders. Task-specific tremors have been defined in the consensus statement on tremor in 1998.¹ The most common task-specific tremor is PWT, which was first described by Rothwell in 1979.² There are two types of PWT: *type A*, which refers to tremor induced by writing, and *type B*, which refers to tremor induced when holding the arm in the position for writing.²

It has since been an ongoing discussion, whether PWT is a type of focal dystonia, a local form of essential tremor, or a nosological entity of its own.^{1,3–6} Recent research suggests that it is rather a separate entity.^{5–10}

Primary Bowing Tremor and PWT

We report on a violinist in his early 60s, who presented to our institute because of a unilateral tremor. The tremor was present in the right arm and induced by playing the violin. Age of onset was 46. He fulfilled the three criteria for focal task-specificity as described by Rosenbaum and Jankovic³: a) tremor was unilateral and limited to one body part, i.e., the right arm (focal); b) no other movement disorders were present; and c) tremor was at first provoked only by playing the violin (task specific), corresponding to a *type A* tremor. It was mainly a pronation-supination tremor, similar to the movement described in PWT by Rothwell.²

Tremor spread to other tasks about 14 years after onset of primary bowing tremor. However, this did not affect his everyday life, and tremor at the violin remained the most prominent and disturbing symptom. Tremor spreading to other tasks has been described in PWT before,^{4,11} and a strict task-specificity was thus negated.^{10,11} Rather, an evolution of the disorder at different points in time¹¹ was discussed.

Family history was positive, with the father suffering from either WC or PWT. This alludes to a genetic component in bowing tremor that has been described in PWT.^{4,11}

Environmental factors may play a role in primary bowing tremor as well as in PWT. Bain et al.⁴ showed that 19% of patients with PWT had a previous history of trauma. This corresponds to our patient, who had been involved in a car accident with a subsequent pain syndrome of the right arm prior to tremor onset.

Our treatment included botulinum toxin, which led to a slight improvement. An improvement of tremor after botulinum toxin injection also has been reported in PWT.¹²

Oral medications included propranolol, primidone, gabapentin and trihexiphenidyl, of which most effective were primidone and propranolol. Trihexiphenidyl had a slight effect. This is in accordance with reports that about 33% of patients with PWT respond specifically to primidone or propranolol.⁴ Alcohol, which also has been reported to improve symptoms in about 33% of PWT-patients,⁴ also relieved tremor in our patient.

CONCLUSION

From a phenomenological point of view, our patient's symptoms are very similar to those displayed in patients with PWT. Both tremors occur in a highly trained fine motor task. We therefore hypothesize that primary bowing tremor (PBT) and PWT may share similar underlying pathophysiological mechanisms so that PBT may be an entity of its own, as has been proposed by Lederman¹³ (2010).

The main pathophysiologic findings in PWT that corroborate the definitions of PWT as an entity of its own are:

1. No excessive overflow in EMG recordings in PWT as opposed to WC.^{4,8}
2. Normal reciprocal inhibition in PWT^{4,8} as compared to WC.^{14,15}
3. In a recent study by Meunier et al.,¹⁶ TENS was shown to have a deteriorative effect in PWT⁶, whereas in WC high-frequency TENS may be beneficial.
4. Normal results for spinal and motor cortex excitability⁸ were obtained in PWT, whereas it may be abnormal in essential tremor and is abnormal in WC.^{17,18}

Defining PBT as an entity of its own, similar to PWT, has the advantage in that it will allow a systematic approach to new hypotheses concerning the pathophysiology of PBT:

1. PBT shows no excessive overflow of EMG activity.
2. PBT show normal reciprocal inhibition.
3. TENS does not have a beneficial effect on PBT.
4. PBT has a normal intracortical and spinal excitability.

With regard to treatment options, we would hypothesize that thalamic stimulation may have a beneficial effect on PBT, as has been shown for PWT.^{19,20} Furthermore, brain imaging in PBT should reveal a distinct activation pattern similar to that in PWT.⁷ Future research is needed to clarify these hypotheses.

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