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Review

# Advances in primary writing tremor☆

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ABSTRACT

Primary writing tremor (PWT) is considered to be a type of task-specific tremor in which tremor predominantly occurs and interferes with handwriting. The pathophysiology of PWT is not clear. Primary writing tremor may be a variant of essential tremor, a type of focal dystonia such as writer's cramp, or a separate nosological entity. Botulinum toxin injections and deep brain stimulation may be treatment choices for primary writing tremor.



**Contents**



## 1. Introduction

Tremor is an involuntary oscillation of a body part and is the most common form of movement disorder. Task-specific tremor is largely or solely limited to a specific task or movement, such as writing, speaking, smiling, or standing [1]. Primary writing tremor (PWT) is considered to be a type of task-specific tremor in which tremor occurs and interferes with handwriting [2–5]. The pathophysiology of PWT is unknown [3,6,7]. There is still some controversy as to whether PWT is a variant of essential tremor [7,8], a type of focal dystonia such as writer's cramp [9], or a separate nosological entity [2,6].

## 2. Clinical manifestation of PWT

First described in 1979 by Rothwell et al. [10], the term PWT is used to define a specific action tremor in which pronation of the forearm elicits a pronation/supination tremor during writing but not seen during other arm movements. They described a right-handed young man who presented with difficulty writing caused by bursts of tremor which occurred whenever the right forearm was pronated. Patients are classified as having either type A or type B PWT depending on whether tremor appears during writing (type A: task induced tremor) or while writing and also on adopting the hand position normally used for writing (type B: positionally

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sensitive tremor) [2]. Examination revealed that the affected hand was tremulous during writing and (in type B cases) when adopting the hand position normally employed for writing [2]. Bain and colleagues studied twenty one patients with PWT, twenty of whom were male. They explained social bias, for example gender differences in types of employment or the fact that British men retired five years later than women, allowing more time for PWT to appear. The mean age at symptom onset was 50.1 years. During writing tremor, none of the type A cases had progressed and only type B patients had noted any change in their tremor. PWT is usually a relatively static condition and should remain focal; this particularly applies to type A cases; however, there was one patient with bilateral PWT [8].

PWT may occur sporadically or be inherited as an autosomal dominant trait. In Bain's study [2], 33% patients had a family history of PWT, including two brothers with type B writing tremors. The presence of a family history of PWT implies that there is genetic susceptibility to the condition, even so, the incidence of preceding trauma (19%) suggests that the expression of tremor may also depend on environmental factors [2]. No causative gene or mutation is known.

The condition is usually regarded as non-progressive, and some patients have the condition for decades with no further symptoms or remissions. The disability caused by this condition can vary from mild to considerable, depending in the profession of the patient. In Japan where calligraphy is important, thalamotomy has been successfully performed for this condition, as it can threaten the professional career of the patient [4] (Fig. 1).

## 3. Hypothesis

The origin of the entity remains unknown, but that the following findings argue for or against PWT being an essential tremor variant, a type of dystonia, or a separate entity.

### 3.1. Hypothesis 1: PWT is a variant of essential tremor

Jimenez-Jimenez and colleagues [8] reported a 59-year-old man with a 5-year history of a typical primary writing tremor in the right hand who developed similar symptoms in the left hand; he had a sister with essential tremor. He had a good response to alcohol and propranolol. So they suggested that the clinical picture of PWT in this patient was likely associated with essential tremor.

Forty one percent of the patients who drank alcohol found their writing tremor improved after 2 units. This is a similar rate of responsiveness to that found in hereditary essential tremor (50%) [2].

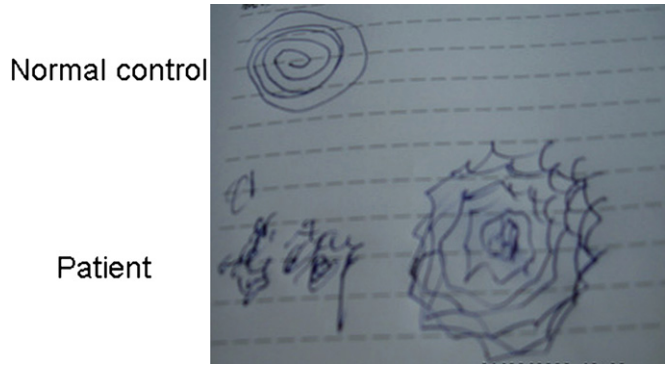


**Fig. 1.** Example of the patient's writing and spirals drawn with the affected hand.

Modugno et al. [7] investigated the excitability of both cortical and spinal motor systems in patients with a definite diagnosis of PWT and compared the results with published data of patients with essential tremor and writer's cramp. They used electrical stimulation of the median and radial nerve to study reciprocal inhibition of forearm antagonist muscles and paired transcranial magnetic stimulation at short and long interstimulus intervals (ISIs) to assess intracortical excitability. The early (presynaptic) and late (disynaptic) phases of reciprocal inhibition were normal as was intracortical excitability at short and long ISIs. Intracortical and spinal excitability are normal in patients with PWT but are abnormal in writer's cramp. In some patients with essential tremor spinal excitability can be abnormal. The study suggests that the pathophysiology of PWT is different from that of writer's cramp and partially also from that of essential tremor (Table 1).

### 3.2. Hypothesis 2: PWT is a type of focal dystonia

Soland et al. [9] reported that nine patients with focal task-specific tremor had neither a significant postural tremor nor a family history. They suggested that focal task-specific tremors were perhaps forms of focal dystonia. The presence of mild dystonic posturing, and response to anticholinergic drugs in some cases of PWT, favors the hypothesis. Singer et al. [11] described a PWT patient with successful treatment with botulinum toxin type A injections, and addressed a possible underlying dystonic mechanism.

Transcranial magnetic stimulation is used over the motor cortex with the conditioning stimulus and the testing stimulus. With this technique, a subthreshold conditioning shock is given before a suprathreshold test shock. This is cortical and suggests that it reflects activity of local intracortical inhibitory (probably GABAergic) interneurons. The time course of paired-pulse testing is complex, beginning with a period of pronounced suppression followed by less prominent facilitation. Less suppression early in the time course indicates that there is a relative decrease in the excitability of cortical inhibitory systems. In writer's cramp there is

**Table 1**
The clinical characteristics of the two types of primary writing tremor and writer's cramp.

| Clinical characteristics | Primary writing tremor | | Writer's cramp |
|---|---|---|---|
| | Type A | Type B | |
| Writing difficulties | Yes | Yes | Yes |
| Other movements | Normal | Normal | Normal |
| Tremor | Task induced tremor | Positionally sensitive tremor | Task induced abnormal posturing and tremor or no tremor |
| Examination | Tremulous during writing | Tremulous whilst adopting the hand position normally employed for writing | Abnormal posturing |
| Static condition | Yes | Yes | Progressed |
| EMG | No excessive overflow | No excessive overflow | Overflow |
| Reciprocal inhibition | Normal | Normal | No H-reflex |
| Disynaptic and presynaptic inhibition | Normal | Normal | Reduced presynaptic inhibition |
| Tremor bursts on tendon taps to the volar aspect of the dominant wrist | No | Yes | – |