

Resting and Reemergent Tongue Tremor as Presenting Symptoms of Parkinson's Disease

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A 73-year-old male was referred to the movement disorders clinic reporting a 12-month history of tongue tremor, which was initially intermittent, becoming more continuous and troublesome with time. A few months later, a right-hand rest tremor appeared along with micrographia and gait difficulties.

The patient had a medical history of diabetes mellitus and prostatic surgery. His family history disclosed a nephew with a diagnosis of Parkinson's disease (PD) at the age of 49 (negative for G2019S/R1441H LRRK2 mutations and GBA screening of pathological mutations).

Neurological examination disclosed tongue rest tremor inducing a slight lip tremor reemerging after approximately 5 seconds of the tongue lying at rest in the floor of the mouth or after opening the mouth; this was accompanied by asymmetric bradykinesia and rigidity of the upper limbs (right > left) and shuffling gait. No history of rapid eye movement sleep behavior disorder or olfactory complaints were reported. Mini-Mental State Examination score was 27. Cerebral CT was normal. No red flag symptoms or exclusion criteria for PD were outlined, and a diagnosis of clinically probable PD¹ was made. The patient started treatment with levodopa/carbidopa 25/100 half tab three times a day (TID) with moderate improvement of hand tremor, but tongue tremor persisted unmodified (see Video 1, Segment A). L-dopa treatment was subsequently increased to 25/100 tid and complete remission of tongue tremor (see Video 1, Segment B) and gait improvement were achieved, although a grade 2 upper limb bradykinesia was still present (MDS-UPDRS-III 19; H & Y stage 2). Written and signed informed consent was obtained from the patient for publication of this case report and related video.

We describe the case of a PD patient whose initial and troublesome symptom was a tongue tremor that subsequently presented an optimal response to 300 mg/day of L-dopa.

In the past, isolated tongue tremor has been reported in patients with essential tremor (ET), with a frequency of 3.8% and generally affecting up to 30% of ET patients if associated with tremor of another body part.^{2,3} However, tongue tremor in ET patients is usually associated with neck, voice, chin, and, more frequently, upper limb tremor. For this reason, in 2008, a classification of orolingual tremor was proposed, suggesting that classical essential orolingual tremor should be diagnosed in the presence of a 4- to 10-Hz tremor, affecting the orolingual structure, present mainly on action (including speaking and swallowing), accentuated by some tasks, but not task specific and associated with ET of the upper limbs occurring at or before onset of the orolingual tremor, in the absence of other neurological signs.³ Therefore, isolated tongue tremor has been excluded. Tongue tremor is also generally accepted as a common sign of PD patients⁴ if accompanied by other parkinsonian signs whose presence fulfill the diagnosis of PD according the UK Brain Bank criteria.³ However, very few reports describe its features, probably because it is usually not considered bothersome by patients or investigators. There are no systematic data on the frequency of rest tongue tremor among PD patients, and specific response to treatment has been reported. A single report on reemergent tongue tremor as an initial presentation of PD and good response to a combined therapy with pramipexole (3 mg/day) and L-dopa (150 mg/day) has been recently published.⁵ Taking into account all these data, even if essential tongue tremor is mainly present on action, possibly accentuated by tasks when compared to the rest tongue tremor of PD patients,

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there are no definitive clinical features that can clearly differentiate tongue tremor in ET and PD patients, and its diagnosis is probably based more on the associated clinical signs rather than on the characteristic of tongue tremor per se.

Whether or not the tongue tremor of our patient can be defined “reemergent” is speculation as it “reemerges” after approximately 5 seconds of lying at rest in the floor of the mouth, and while parkinsonian reemergent tremor appears after a similar latency of 9.37 (standard deviation: 10.66) seconds in a postural position, presenting the same frequency of rest tremor.⁶

Our case report highlights a likely underestimated and forgotten clinical sign whose presence should induce neurologists to search for other parkinsonian signs and suggests that parkinsonian tongue tremor presents a good response to L-dopa.

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 of the First Draft, B. Review and Critique.

M.F.: 1A, 1C, 3A

L.A.: 1C, 2C, 3B

T.S.: 1B, 3B

J.J.F.: 1A, 1B, 3B

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Supporting Information

A video accompanying this article is available in the supporting information here.

Video 1. Segment A: A rest tremor of the tongue, that “reemerges” after approximately 5 seconds of the tongue lying at rest in the floor of the mouth or after opening the mouth. Segment B: Tongue rest tremor remission under 300 mg/day of L-dopa treatment.