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LETTER TO THE EDITOR

Pseudo-orthostatic tremor in idiopathic Parkinson's disease: could it be re-emergent tremor?

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Dear Editor,

Re-emergent tremor is one of the parkinsonian rest tremors characterized by development within a few seconds of latency after assuming posturing [1]. It occurs predominantly in the upper extremities; however, other body parts with re-emergent tremor have been occasionally reported [2, 3]. Re-emergent tremor differs from essential tremor (ET) in terms of latency, amplitude, and frequency [1]. Kinetic leg tremor can be seen in ET patients, but resting leg tremor is exclusively observed in patients with Parkinson's disease (PD) [4]. In addition, orthostatic or pseudo-orthostatic tremors have also been reported in patients with PD [5, 6]. However, re-emergence of leg tremor during standing has not been demonstrated in patients with PD.

Herein, we report a case of pseudo-orthostatic tremor with delayed latency in PD.

A 48-year-old male patient complained of uncontrollable limb tremor and tremulousness of legs during standing. His symptoms began 3 years prior to evaluation in our clinic. Initially, he presented with crossed limb rest tremor of the left hand and the right foot. The tremor progressed to involve both sides, and bradykinesia and rigidity also developed. One year after the onset of parkinsonism, the patient was diagnosed as having PD in a different neurology clinic. A dopamine agonist, anticholinergic agent, propranolol, and clonazepam were administered with very mild improvement during the subsequent year. The patient had no family history of tremor or parkinsonism. He had been treated for essential hypertension with a combination of antihypertensive agents: 160 mg of valsartan and 5 mg of amlodipine. He did not take any medicine known to cause parkinsonism or tremor, such as prokinetics and neuroleptics, or any other medications other than the aforementioned drugs.

Neurologic examination revealed asymmetric resting tremors which were severe in the left arm and the right leg (video segment 1). Postural or re-emergent tremor was not observed with the arms outstretched. There was no ataxia, intention tremor, or dystonia. He had a subtle bradykinesia and rigidity with cogwheeling. The cranial nerves were normal, and no evidence of a pyramidal or sensory disturbance was found. His functional status was scored two with the modified Hoehn and Yahr scale and 13 on the Unified Parkinson's Disease Rating Scale part 3.

During standing, he did not complain of shakiness at the beginning. Orthostatic tremor, predominantly in right leg, was found after about 8 s which was compatible with the timing of his complaint of unstableness. The delayed onset tremor appeared to have approximately 4–5 Hz of frequency with asynchronous antagonistic muscles activation. The tremors became more intensified, and after about 15 s, his whole body shakiness was noticeable as tremulousness. His left hand began to shake when the body shakiness intensified, and the whole body and left hand tremor intensified together until plateau (video segment 2). The tremulousness of the legs and trunk immediately disappeared after walking, sitting, or lying. The same pattern of delayed onset tremor reiterated after motionless stance.

Surface electromyography was performed on the leg and hand muscles and showed rest tremor which measured 4–6 Hz of frequency and re-emergent leg tremor with a frequency of 4 Hz (Fig. 1). The blood tests, including a thyroid function test, were normal. Positron emission tomography using ^{18}F-N-(3-fluoropropyl)-2beta-carbon ethoxy-3beta-(4-iodophenyl) nortropane demonstrated asymmetric decreased dopamine transporter uptake at the posterior putamen and caudate, mainly on the left side (Fig. 2).

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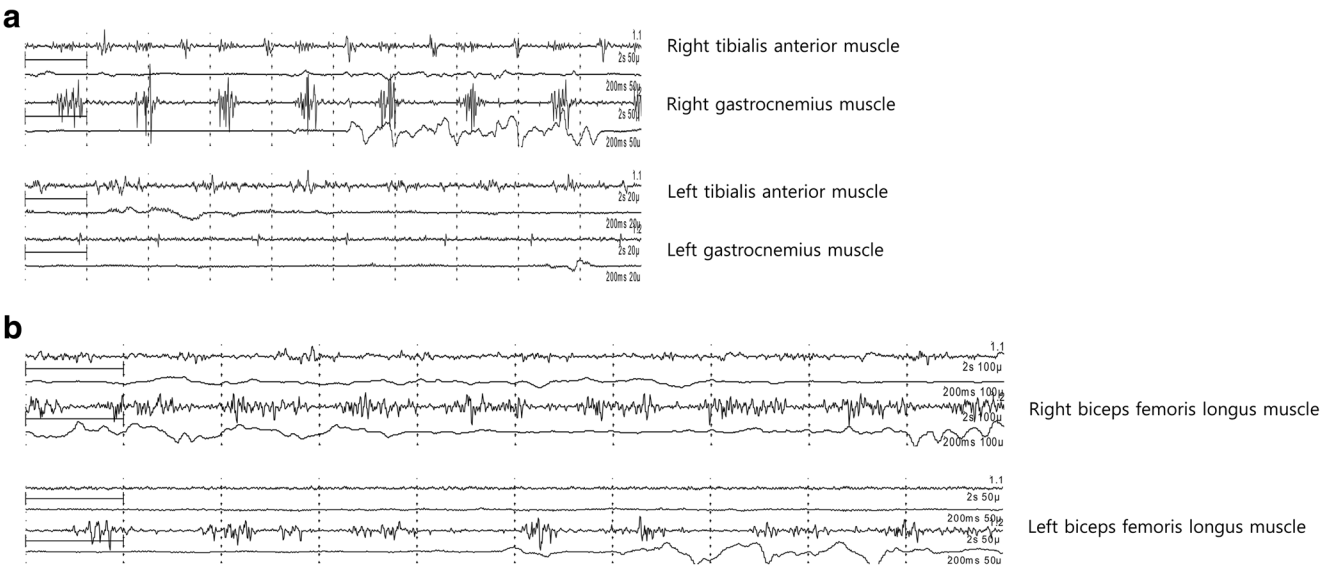

Fig. 1 Two-channel surface electromyography was performed on the leg muscles during sitting (**a**) and standing (**b**) and showed 4- to 5-Hz alternating rhythmic spontaneous motor unit potentials

Primary orthostatic tremor is a rare movement disorders characterized by a tremor of high frequency of 13–18 Hz in the legs that exclusively occurs shortly after standing. Its latency is reported to be less than 1000 ms [7]. This tremor almost completely disappears by sitting or walking. Orthostatic tremor can be idiopathic or secondary, and it can co-occur with other neurological diseases, such as ET or PD [8]. Some PD patients also can have "pseudo-orthostatic" tremor which characterized by a frequency of the 4- to 7-Hz range [5].

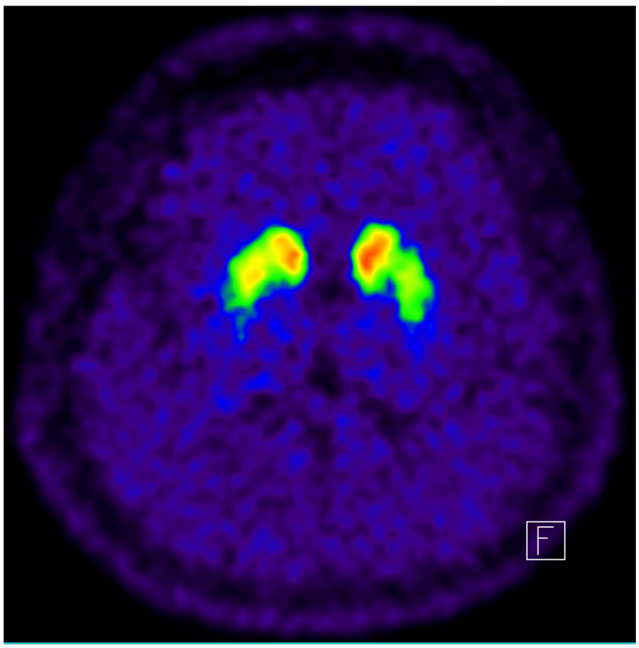

Fig. 2 Positron emission tomography using ^{18}F-N-(3-fluoropropyl)-2beta-carbon ethoxy-3beta-(4-iodophenyl) nortropane demonstrated asymmetric decreased dopamine transporter uptake at the posterior putamen and caudate, and it was most severe in the left

However, to the best of our knowledge, none of the published research mentions latency of orthostatic tremor in PD patients. Therefore, the term re-emergent orthostatic tremor was never used. Phenomenologically, standing can be a counterpart of outstretched arms. Therefore, our patient manifested re-emergent orthostatic tremor with long latency to onset of tremor after standing [1].

Our patient demonstrated many characteristics which suggest re-emergent pseudo-orthostatic tremor is a part of PD tremor. The patient's orthostatic tremor frequency matched that of other body parts [5]. Parkinsonian tremor, whether resting or re-emergent, has a specific range of frequency at 4–6 Hz. In addition, unlike essential tremor and primary orthostatic tremor which show immediate onset [1, 7], our patient's tremor shares re-emergence after posturing as seen in PD [1]. Re-emergent tremor is postulated to occur by transiently interrupting central oscillators, such as the ventrolateral thalamus and pallidus, with proprioceptive sensory information provoked by repositioning of the limbs. These central generators are sensitive to sensory feedback; thus, desynchronizing thalamic or pallidal discharges accounts for re-emergence [1, 9]. Standing provided the equivalent sensory information that disrupted the central oscillator activity as in arm posturing, thus causing the latency in this patient. The simultaneous occurrence of other parkinsonian signs and evidence of decreased dopamine transporter uptake further support the shared origin of this orthostatic re-emergent tremor.

Our patient's hand tremor intensified during standing as the standing leg tremor also intensified. This temporal synchronization of hand tremor with orthostatic leg tremor further substantiates our hypothesis that the same mechanism was shared between the two tremors; the disease pathophysiology was expanding its extent to the topography of the hands within the central generator circuitry [10].

In summary, our patient manifested a re-emergent orthostatic tremor that is a part of PD symptomatology.

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Compliance with ethical standards

Ethics The institutional review board at St. Mary's Hospital approved this case report.

Conflict of interest The authors declare that they have no conflict of interest.

Patient consent The patients have consented to the submission of the case report to the journal.

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