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Unilateral elbow joint festination in early Parkinson's disease

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Festination is a common and disabling gait disturbance associated with Parkinson's disease (PD) and other Parkinsonian syndromes. Although festination predominantly affects gait, it can also affect non-gait motor systems [1]. However, unilateral elbow motion festination in early Parkinson's disease without gait festination has never been reported.

A 60-year-old right-handed male diagnosed with PD 3 years prior to presentation noticed difficulties while tooth brushing in terms of keeping pace and difficulties stopping his tooth brushing. He complained of increasingly rapid and bigger flexion-extension movements of the elbow joint that often subsequently hurt his mouth. Needle electromyographic recording of biceps and triceps muscles shows an increase in the frequency (up to 6–7 Hz) and amplitude without sequence effect or involuntary stop (Supplemental Fig. 1A). These uncontrolled movements occurred when he did any movement using the elbow joint regardless of holding a toothbrush and did not occur with his right shoulder movement or with contralateral upper extremity movement. (Supplemental Video 1). Slowing the speed of the elbow movement (less than one stroke per 1 sec) ameliorated his symptom but auditory sensory tricks (hearing the metronome, hand clapping with slow and regular rate) did not any help. The patient did well with other complex slow movements that involve the elbow joint such as using chopsticks, cutting oranges with a knife or coin rotation test. Additionally, there was no ideomotor or ideational apraxia.

Video segment demonstrates increasingly rapid and bigger movements of the elbow joint during tooth brushing. (segment 1) These uncontrolled movements occurred when he did any movement using the elbow joint and did not occur with his right shoulder movement (segment 2) Patient gave consent to be videoed for publication.

Supplementary video related to this article can be found at https://doi.org/10.1016/j.parkreldis.2019.06.016.

A neurological examination revealed left side-dominant rigidity, bradykinesia and 6–7 Hz resting hand tremors (Supplemental Fig. 1B) without gait freezing or festination. He was in HY stage II, motor UPDRS III 22, with MMSE 29/30 without wearing off and dyskinesias on 300 mg levodopa and 1.5 mg of Pramipexole. Medications (levodopa, amantadine, propranolol and anticholinergics) did not ameliorate his symptoms and the patient had to begin using an electric toothbrush. A brain magnetic resonance imaging (MRI) scan was normal but a FP-CIT positron emission tomography (PET) scan revealed decreased uptake in the bilateral posterior putamen with more affected on the right side(Supplemental Fig. 2A). He exhibited reduced cardiac cardiac meta-iodobenzylguanidine (MIBG) level, suggesting idiopathic PD (Supplemental Fig. 2B). Over 3 years of follow-up, the patient did not develop gait freezing or festination and could walk independently.

Festination is clinically defined as the tendency to speed up and lose normal amplitude during quick, repetitive movements [2]. Thus, elbow joint movement of the present case was compatible with festination. Although festination and freezing predominantly affect the lower limbs, recent reports have shown that festination and freezing can occur during upper [2–8] and lower [9–14] limb movements and speech [15–17]other than gait (Table 1). Dopaminergic deficit in basal ganglia could be involved in timing and amplitude generation of self-paced repetitive movements. Tooth brushing also requires the generation of a cyclic rhythm with an individual's frequency in the elbow joint [18]. Freezing is often preceded by an episode of festination with similar spatiotemporal dysfunction [11], suggesting a shared common mechanism between the freezing and festination. However, in present case, there was no subsequent development of gait festination or freezing over the 3-year follow-up period, suggesting this represent a different entity that is not related to the dopaminergic system. This is in line with recent study that freezing of upper limb was not correlated with that of lower limb and did not decrease after levodopa, suggesting that different brain region involvement lead to different types of freezing [6]. It is also possible that very subtle freezing following festination was not recognized, or the patient was able to compensate the spatiotemporal dysfunction before the development of freezing [19]. From a phenomenological perspective, this movement resembles a form of tremor because there was no sequence effect, poor response to levodopa, and maximal frequencies observed during festination did not exceed the resting tremor frequency (6–7 Hz). A link of festination with the frequency of the tremor has been suggested [20]. However, it is less likely that this phenomenon resulted from impaired dexterity or apraxia because he can perform tooth brushing at a slow speed and can also easily accomplish more complex movements using the elbow joint. Similar cases in PD or healthy controls will be needed to understand the pathophysiology of festination of elbow joint.

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Table 1
Summary of freezing and festination of upper and lower limb and speech other than gait in PD.

Body part	Motor Task	Motor phenomenon	Reference
Speech	Reading	Festination, freezing	Cantiniaux, 2010 Park, 2013
	Orofacial diadochokinetic task	Oral festination	Moreau, 2007
Upper limb	Hand/Finger tapping	Freezing, festination	Bronte, 2000, Yahalom 2004 Stegemöller 2009 Barbe 2014 Delval 2016
	Hand drawing/pointing/ writing	Freezing	Nieuwboer 2009 Almeida 2002
	Piano-playing	Festination, freezing	Rossi 2015
	Elbow joint movement	Festination	Current study
Lower limb	Foot-tapping	Festination, freezing	Nantel 2011 Vercruysse 2012
	Pedaling (including virtual reality)	Freezing	Abe 2003 Gliat 2013 Matar 2013 Shine 2013

Relevant conflicts of interests/financial disclosures

The authors have nothing to declare.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.parkreldis.2019.06.016.

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