Tongue tremor as a manifestation of atypical Parkinsonism treated with coaxial deep brain stimulation of thalamus and subthalamic area

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Tongue tremor is a rare, initial presenting symptom of Parkinson’s disease [1] and other Parkinsonism conditions. The tongue tremor may be medically refractory [1,2]. The therapeutic role of deep brain stimulation (DBS) in treating this symptom remains unknown [3]. Here, we describe a patient who presented with tongue tremor as a manifestation of atypical Parkinsonism. We successfully treated him using coaxial thalamic and subthalamic DBS.

A 64-year-old man, without prior use of neuroleptics or medical family history, was referred for tongue tremor that was present for 2 years. The tremor appeared when he was awake and had bothered him significantly. He had hypertension and diabetes mellitus, but did not suffer stroke. The patient had been treated with pranolol, clonazepam, and trihexyphenidyl, with no improvement. Botulinum toxin injection to the tongue was ineffective. His spouse had noticed that his posture and gait had changed a few months prior to his referral. Hyposmia, sleep disorders, and depression were absent.

Upon examination, the tongue showed symmetric to-and-fro and up-and-down motions of 3–4 Hz at rest, like a butterfly’s wings’ flutter (Video). The tremor disappeared with voicing and protrusion but reemerged immediately (i.e., re-emergence phenomenon [1]). Associated tremors were seen in the soft palate and lower face, but not in other body parts. His Fahn-Tolosa-Marin tremor rating scale (TRS) score was 7. In addition, his speech was monotonous. Diminution of facial expression, limb rigidity, reduction of leg agility, stooped posture, and postural instability were mild. His gait and action when rising from a chair were slow. His score on the unified Parkinson’s disease rating scale (UPDRS) part III was 18. The rest of the neurological examination was normal.

The video shows preoperative and postoperative tongue tremors with ‘on’ and ‘off’ DBS.

Supplementary video related to this article can be found at http://dx.doi.org/10.1016/j.parkreldis.2017.08.023.
therapeutic effects with minimum adverse effects, by combining multiple active cathodes and anodes. In the present case, the most distal electrode was used for additive PSA simulation, which was effective for other Parkinsonism symptoms, as observed in patients with Parkinson’s disease [5]. Coaxial stimulation of the thalamus and subthalamic area may be a useful option for tremor-dominant Parkinsonism symptoms.

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Fig. 1. A: Preoperative magnetic resonance (MR) imaging showed normal anatomy of the inferior olivary nuclei (square). B: Dopamine transport scintigraphy-123I-Ioflupane imaging showed reduction of uptake in the bilateral striatum (square). C, D: The targets of the deep brain stimulation (arrowheads) on the thalamus (C) and subthalamus (D), plotted on T2-weighted MR images. The red nucleus is marked using a dotted circle. E: Microelectrode recordings of the thalamic ventral tiers showed activity of the kinetic cells in the ventral intermediate nucleus (top). The background amplitude of the recording slightly decreased in the subthalamic area (bottom). F: Bilateral thalamic electrodes (arrowheads) on postoperative fused images of computed tomography-MR. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

References

Hiroki Toda
Department of Neurosurgery, Tazuke Kofukai Medical Research Institute and Kitano Hospital, Osaka, Japan

Kotaro Asanuma
Department of Neurosurgery, Fukui Red Cross Hospital, Fukui, Japan

Takayuki Kondo
Department of Neurology, Yanagibaba Takeda Clinic, Kyoto, Japan

Yuuta Terada
Department of Neurology, Tazuke Kofukai Medical Research Institute and Kitano Hospital, Osaka, Japan

Hidemoto Saiki
Department of Neurology, Tazuke Kofukai Medical Research Institute and Kitano Hospital, Osaka, Japan

* Corresponding author. Department of Neurosurgery, Tazuke Kofukai Medical Research Institute and Kitano Hospital, 2-4-20, Ohgimachi, Kita, Osaka, 530-8480, Japan.
E-mail address: htoda-nsu@umin.ac.jp (H. Toda).

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