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Case Report

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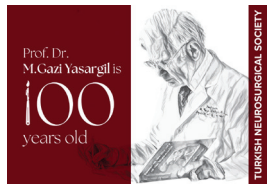

A Case of Tongue Twisting During Screening of STN-DBS for Parkinson's Disease: A Unique Form of Pyramidal Tract Activation

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ABSTRACT

Subthalamic nucleus deep brain stimulation (STN-DBS) is a safe and effective therapy for Parkinson´s disease (PD) in selected patients. However, various side effects such as paraesthesia, diplopia, ataxia, worsened akinesia, emotional changes, dysarthria, and muscle contractions can occur due to the current spread to the adjacent structures during the STN-DBS programming sessions. Muscle contractions result from the corticospinal and corticobulbar side effects, which can manifest due to the current spread to the pyramidal tract during DBS programming. Here, we report a case of tongue-twisting movement as a unique corticobulbar side effect of the STN-DBS programming in a patient with PD.

KEYWORDS: Parkinson's disease, STN-DBS, Corticobulbar side effects

INTRODUCTION

Subthalamic nucleus deep brain stimulation (STN-DBS) is an effective and well-tolerated therapeutic option for carefully selected patients with Parkinson´s disease (PD). However, various side effects can occur, some resulting from the spread of electrical current to adjacent structures, such as the pyramidal tract (4,7). Corticospinal and corticobulbar side effects include muscle contractions, spasms, and gait and speech problems (7). In this report, we present a patient with PD who developed a tongue-twisting movement as a unique side effect of STN-DBS affecting the corticobulbar tract.

CASE REPORT

A 38-year-old woman presented with shoulder pain, stiffness, flexed posturing of the right arm, and dragging of the right foot. She was diagnosed with idiopathic PD. By age 43, she experienced motor fluctuations, left leg foot posturing with inward turning, intermittent dyskinesias mainly affecting her head and neck, and mood disturbance. Her medications consisted of 62.5 mg of levodopa plus benserazide three times per day, 100 mg of amantadine twice daily, and 10 mg of ropinirole once daily. Laboratory evaluations of serum copper and ceruloplasmin were within normal limits, and genetic testing for parkin, DRPLA, and SCA 1, 2, 3, 6, and 7 gene mutations was negative. Cranial magnetic resonance imaging (MRI) was unremarkable, and DaTscan findings confirmed parkinsonism.

During an L-dopa challenge test prior to surgery, the Unified Parkinson's Disease Rating Scale motor score (UPDRS III)

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was 46 in the off-medication state and 25 in the on-medication state, indicating a 45.6% improvement. Gait and speech were satisfactory, although neuropsychological assessment revealed mild cognitive impairment. STN-DBS was performed under general anaesthesia at age 51 using the Leksell G-frame and an MRI-guided and MRI-verified technique without microelectrode recording. Boston Scientific Cartesia directional leads were implanted bilaterally. Initial DBS settings were contact 4–1.8 mA and contact 10-1.6 mA; 60 µs, 130 Hz. Postoperative medications included 125 mg of levodopa plus benserazide five times per day and 100 mg of amantadine twice daily.

Eight months following surgery, right foot dystonia affecting gait was noted. As DBS settings were adjusted to all contacts of level 2 (contacts 2, 3, and 4), 2 mA; 60µs, 130 Hz, the right foot dystonia resolved, but a twisting, turning movement of the tongue occurred, worsening as stimulation increased to 2.5 mA and then to 2.8 mA. The tongue-twisting movement ceased without reoccurrence of foot dystonia, as the stimulation was steered posteriorly and medially. Volume of tissue activated (VTA) modelling using the Boston Scientific GuideXT platform revealed lateral and anterior spread of stimulation, most likely into the corticobulbar tract, associated with the tongue-twisting movement (Figure 1).

Written informed consents were obtained from the individuals (and/or legal representatives) for the publication of the cases.

DISCUSSION

This report describes a tongue-twisting movement observed during STN-DBS screening in a patient with PD. Understanding this involuntary movement requires consideration of the neuroanatomy of tongue innervation, which is crucial for vital functions including respiration, speech, and swallowing.

The tongue receives motor innervation via the hypoglossal (XII) nerve and includes extrinsic (genioglossus, styloglossus, hyoglossus, and palatoglossus) and intrinsic (superior and inferior longitudinal, transverse, and vertical) muscles. Since the tongue lacks internal bones, contractions in its muscles produce three-dimensional changes in shape. The extrinsic

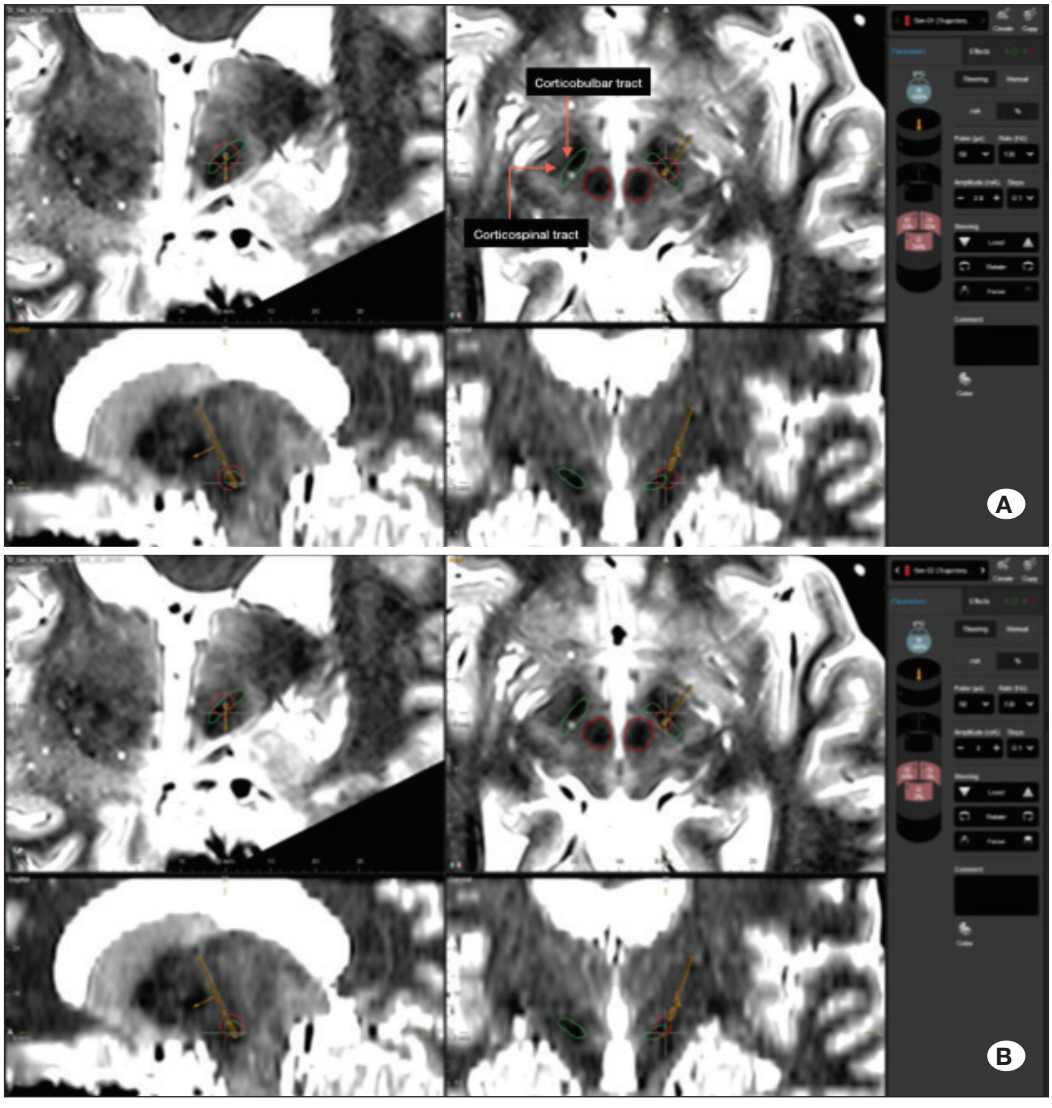

Figure 1: Volume of tissue activated (VTA) modelling using the Boston Scientific GuideXT platform.
A) Lateral and posterior spread of stimulation using ring mode, most likely into the corticobulbar tract, resulting in tongue twisting.
B) Directional stimulation steered posteriorly and medially, resulting in the resolution of the tongue-twisting movement.

muscles primarily control protrusion, retraction, and side-to-side movement, while the intrinsic muscles are more likely to alter tongue shape, producing movements such as rolling, twisting, curling, uncurling, flattening, and rounding of its surface (5). However, there are still knowledge gaps in understanding the exact role of each muscle in overall tongue shape and movement (9).

Motor innervation of the tongue originates in the precentral gyrus and other frontal cortical regions and travels via corticobulbar fibres through the corona radiata, genu of the internal capsule (IC), cerebral peduncle, basis pontis, and medullary pyramid, with bilateral connections to the hypoglossal nuclei (8). Although there is limited knowledge about the somatotropic organisation of axon fibres in the IC, several reports suggest that orofacial symptoms may occur due to involvement of the genu and posterior limb of the IC (2,10).

While the corticobulbar fibres have traditionally been considered to descend through the genu of the IC, Brissaud reported that some fibres in the genu could lead to voluntary movements of the face and tongue (3). However, Bertrand showed that regions related to facial movements were more likely located in the posterior limb of the IC rather than the genu (2). Supporting this, Yim et al. reported that corticobulbar tracts originating from the precentral gyrus localised in the middle and third portions of the posterior limb of the IC rather than the genu. Thus, corticobulbar tract fibres are thought to be positioned around the midpoint of the posterior limb of the IC (11).

From an anatomical point of view, during STN-DBS, especially at higher stimulation settings, the lateral spread of current into the corticospinal and corticobulbar tracts, known as the pyramidal tract, located around the posterior limb of the IC, is considered responsible for several challenging side effects, including muscle contractions, twitches, gait and speech disturbances, and pain (1,6,7,10).

CONCLUSION

We observed an isolated tongue-twisting movement in our patient during STN-DBS screening for PD. This occurred while stimulating contact 2 and worsened as stimulation amplitude increased (Figure 1A). As the tongue-twisting movement ceased when stimulation was directed medially (Figure 1B), we considered this a unique capsular effect of STN-DBS.

Patient confidentiality and consent document has been obtained.

Declarations

Funding: P.L. has received honoraria and travel expenses from Boston Scientific and Medtronic for speaking at meetings. L.Z. is a consultant for Boston Scientific and Medtronic.

Availability of data and materials: The datasets generated and/or analyzed during the current study are available from the corresponding author by reasonable request.

Disclosure: The authors declare no competing interests.

AUTHORSHIP CONTRIBUTION

Study conception and design: YD, PL, VD, HA
Data collection: YD, VD, HA
Analysis and interpretation of results: PL, MH, HA, VD, LZ
Draft manuscript preparation: YD, VD, HA
Critical revision of the article: MH, PL, LZ, HA
Other (study supervision, fundings, materials, etc...): PL, MH, LZ
All authors (MH, PL, LZ, HA, VD, YD) reviewed the results and approved the final version of the manuscript.

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