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Case Report

Radiofrequency ablation of the pallidothalamic tract and ventral intermediate nucleus for dystonic tremor through the parietal approach

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ABSTRACT

Background: The thalamic ventral intermediate nucleus (Vim) and globus pallidus internus are far apart and cannot be captured using a single electrode.

Case Description: We describe our experience with a patient with dystonic tremors of the head and upper and lower extremities who showed symptomatic improvement after radiofrequency (RF) ablation using a parietal lobe approach with a single trajectory to capture the pallidothalamic tract and Vim. A 46-year-old man developed head tremors at 41 and a right-sided neck tilt three years later. Five years after the onset of the head tremors, tightness of the larynx during speech and tremors in both the upper and lower limbs also appeared. The Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) score was 24, and the Fahn-Tolosa-Marin Tremor Rating Scale (FTM) score was 48. We captured the pallidothalamic tract and Vim along a single trajectory by locating the entry point in the inferior parietal lobule. One week after treatment, the TWSTRS and FTM scale scores were 9 (62.5%) and 30 (37.5%), respectively. No adverse events were observed.

Conclusion: This case suggests that in dystonic tremors involving abnormalities of the basal ganglia-thalamo-cortical and cerebello-thalamo-cortical circuits, a single electrode can be used to approach both circuits through the parietal lobe approach.

Keywords: Dystonic tremor, Pallidothalamic tract, Parietal lobe, Ventral intermediate nucleus

INTRODUCTION

The optimal target for the stereotactic neurosurgical treatment of dystonic tremors has not yet been established. Deep brain stimulation (DBS) of the ventral intermediate nucleus (Vim) is the preferred surgical treatment for dystonic tremors.[13,10] However, Vim-DBS is sometimes insufficient to improve dystonia symptoms. While globus pallidus internus (GPi)-DBS can also be used to treat dystonic tremors,[13] it provides insufficient control of tremor symptoms. Recent studies have reported the efficacy of combined Vim and GPi DBS for treating dystonic tremors.[10,12] The thalamic Vim nucleus and GPi are far apart and cannot be captured using a single electrode. Therefore, the implantation of one DBS electrode each for the GPi and Vim is required. We previously reported that RF ablation of the pallidothalamic tract (PTT), consisting of the output fibers of the GPi, can improve dystonia similarly to RF ablation of the GPi

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(pallidotomy).[6,8] We describe our experience with a patient with dystonic head and upper and lower extremities tremors who showed symptomatic improvement after RF ablation using a parietal lobe approach with a single trajectory to capture the PTT and Vim. In dystonic tremors involving abnormalities of the basal ganglia-thalamo-cortical and cerebello-thalamo-cortical circuits, a single electrode can be used to approach both circuits through the parietal lobe approach.

CASE REPORT

A 46-year-old man developed head tremors at 41 and a right-sided neck tilt three years later. Five years after the onset of the head tremors, tightness of the larynx during speech and tremors in both the upper and lower limbs also appeared. Because the symptoms were refractory to oral medications, the patient was referred to our department for surgical treatment. He had no history of head trauma, treatment for psychiatric disorders, or a family history of movement disorders. Magnetic resonance imaging (MRI) of the head revealed no apparent structural abnormalities. The patient's symptoms included right lateral flexion and forward neck bending, horizontal head tremors, and tremors in both the upper and lower extremities [Video 1]. Dystonic tremors were diagnosed based on the presence of the tremor and dystonia symptoms in the cervical region. The Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) score was 24, and the Fahn-Tolosa-Marin tremor rating scale (FTM) score was 48. We suggested DBS; however, the patient refused to receive an implanted instrument. Due to his strong desire to improve the tremor in the right upper extremity and improve cervical symptoms, we decided to treat him with RF thermocoagulation of the thalamic Vim nucleus and PTT.

BrainLab elements were used for the surgical planning. The targets were the left PTT for cervical dystonia due to the right-sided neck flexion and the left Vim nucleus for the right upper extremity tremor. The electrode tip for the left PTT was set at 8 mm lateral, 0.5 mm posterior, and 3 mm inferior to the midpoint of the anterior commissure-posterior commissure. We captured the PTT and Vim along a single trajectory by locating the entry point in the inferior parietal lobule [Figures 1 and 2]. T1/T2-weighted thin-slice MRI was performed using a Leksell G frame for surgical planning. A Leksell neurogenerator (Elekta, Stockholm, Sweden) and monopolar RF probe (1.0-mm-diameter tip with a 4.0-mm uninsulated length) were used for stimulation and coagulation. Surgery was performed under local anesthesia in the semi-sitting position without microelectrode recording or impedance monitoring. The bottom of the Vim nucleus was located 9 mm above the target PTT along the trajectory. The coordinate of the Vim nucleus was 15 mm lateral, 6 mm anterior, and 2 mm superior to the posterior commissure.

Video 1: Preoperative condition.

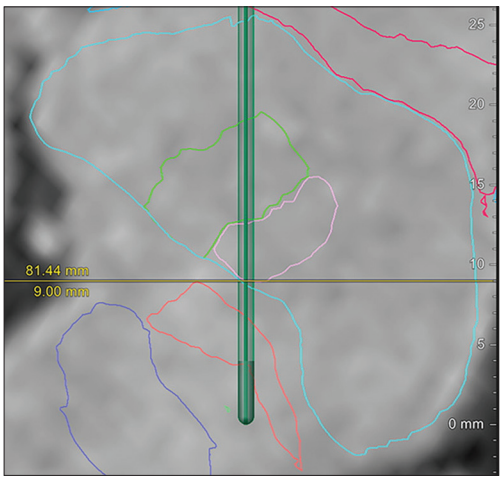

Figure 1: Planned trajectory for the pallidothalamic tract (PTT) and ventral intermediate nucleus (Vim) in Brainlab Elements. The tip of the electrode is set to the left PTT. The bottom of the Vim is positioned 9 mm above the electrode tip.

Macrostimulation was performed on the bottom of the Vim. A1.5 mA/100 μs/133 Hz stimulation caused numbness in the right side of the patient's mouth. When the electrode was moved 1 mm deeper, stimulation was performed again, with no symptoms of numbness. As the tremor symptom of the right upper extremity was alleviated, thermal coagulation was performed at this site at 70°C for 30 s. The tremor in the right upper extremity disappeared [Video 2]. The electrode was advanced to the PTT target, and macrostimulation was performed with no apparent adverse events. Thermal coagulation was performed at the PTT at 70°for 40 s. No evident complications were observed.

Postoperative MRI revealed lesions in the PTT and Vim nuclei, as per the preoperative surgical plan [Figure 3]. While the cervical dystonia improved, symptoms during vocalization and tremors in the right lower extremity did not. One week after treatment, the TWSTRS and FTM scale scores were 9 (62.5%) and 30 (37.5%), respectively [Video 3]. No

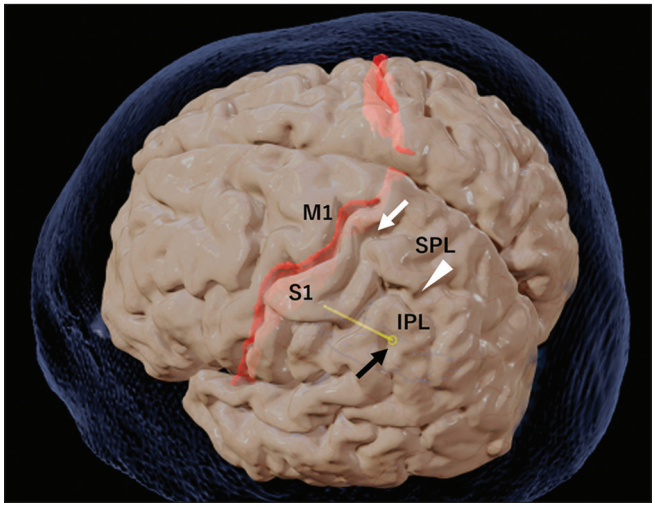

Figure 2: Entry point of the electrode on the brain surface. Black arrow: Electrode (yellow) entry point on the brain surface. White arrow: Post-central sulcus. White arrowhead: Intra-parietal sulcus. The electrode entry point is at the inferior parietal lobule (IPL). M1: Primary motor cortex; S1: Primary somatosensory cortex; SPL: Superior parietal lobule.

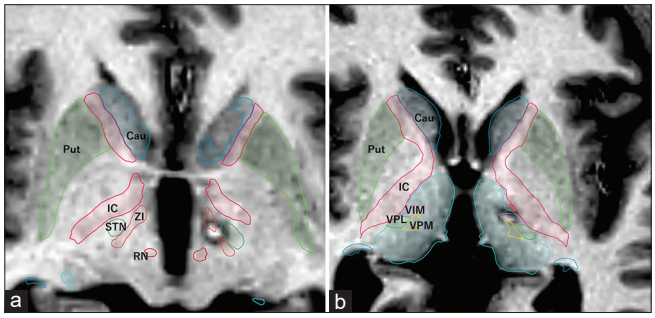

Figure 3: Postoperative T_1-weighted MRI at the PTT and Vim nucleus level. (a) Lesion of the left PTT located medial to the STN and anterior to the RN. (b) Lesion of the left Vim nucleus located within the Vim nucleus, as simulated by anatomical mapping of the BrainLab elements. PTT: Pallidothalamic tract, MRI: Magnetic resonance imaging, Cau: Caudate nucleus, Put: putamen, IC: Internal capsule, RN: Red nucleus, STN: Subthalamic nucleus, Vim: Ventral intermediate, VPL: Ventroposterolateral nucleus, VPM: Ventroposteromedial nucleus, ZI: Zona incerta.

adverse events were observed. Evaluation at six months after surgery showed no significant change from the condition at one week.

DISCUSSION

We applied the parietal approach to capture the PTT and Vim in a single trajectory, and RF thermocoagulation of the PTT and Vim improved cervical dystonia and tremor in the right upper extremity in this case. Using a single trajectory for the basal ganglia-thalamo-cortical and cerebello-thalamo-cortical circuits is a meaningful approach for treating dystonic tremors.

Video 2: Intraoperative condition.

Video 3: One-week postoperative condition.

Studies examining functional connectivity using resting-state functional MRI reported a wider range of functional connectivity abnormalities for dystonic tremors compared to essential tremors.[3,14] A study examining structural and functional connectivity based on the stimulus-effective site of DBS in essential and dystonic tremors also reported that essential tremors were primarily associated with functional abnormalities of the cerebello-thalamo-cortical circuit, whereas dystonic tremors were primarily associated with functional abnormalities of both cerebello-thalamo-cortical and basal ganglia-thalamo-cortical circuits.[14] Based on this background, treating dystonic tremors with simultaneous neuromodulation of the cerebello-thalamo-cortical and basal ganglia-thalamo-cortical circuits is theoretically acceptable. Trompette *et al.* recently reported the results of bilateral combined Vim- and GPi-DBS in three cases of dystonic tremor.[12] In these three cases, three stimulation settings-Vim-DBS only, GPi-DBS only, and Vim/GPi-DBS-were evaluated using the Burke-Fahn-Marsden Dystonia Rating Scale (BFMDRS) and FTM and compared to the off condition.[12] The mean total FTM decreases were 34%, 42%, and 63% for the VIM-only, GPi-only, and combined VIM-GPi-DBS groups, respectively. The mean total BFMDRS decreases were

34%, 37%, and 60% for VIM-only, GPi-only, and combined VIM-GPi-DBS, respectively, compared with the Off condition.[12] Paoli *et al*. also performed the same combined Vim + GPi DBS for tremors with dystonia. They reported a mean improvement of 77.1% in the BFM motor subscore in seven patients at the 2-year follow-up, with significant improvement in tremor symptoms in all but one patient.[10]

Attempts to capture two different circuits with a single electrode have been reported. Buhmann *et al*. performed DBS in three patients with dystonic head tremors.[1] The authors placed the proximal contacts of the electrodes into the inferior base of the ventrolateral (VL) thalamic nuclei, including the ventro-oral (Vo) nucleus, which receives the PTT and the Vim nucleus, and the distal contacts into the posterior subthalamic area (PSA) nucleus, which receives the cerebello-thalamic tract.[1] The optimal stimulation was left PSA and right thalamic VL nucleus stimulation, bilateral VL stimulation, and bilateral PSA stimulation in three patients, with an average 72% improvement in TWSTRS and 63.2% in FTM.[1] Stimulation at the inferior base of the VL nucleus may have stimulated the Vo nucleus, the anterior component of the VL nucleus, and the Vim nucleus, the posterior component of the VL nucleus.

We previously reported the application of Vo-Vim DBS for head tremors by implanting a DBS with a steep angle, in which head tremors disappeared with stimulation of the Vop-Vim border.[15] Stimulation of this site may have stimulated both the pallidothalamic and cerebellothalamic tracts. We found that for cervical dystonia, unilateral PTT resulted in a 46.4% improvement in TWSTRS, whereas unilateral pallidotomy resulted in a 47.9% improvement.[6,8] This suggests that the improvement in cervical dystonia symptoms was comparable between PTT and GPi. While cerebral infarction (4.3%), cerebral hemorrhage (7.2%), Parkinsonism (4.3%), hemiparesis (2.9%), and visual disturbance (1.4%) were confirmed in 69 patients with dystonia after unilateral pallidotomy, reduced hand dexterity (20%), cerebral hemorrhage (2.9%), hypophonia (14.3%), dysarthria (11.4%), and executive dysfunction (2.9%) were confirmed in 35 patients with dystonia after unilateral pallidothalamic tractotomy. Dysarthria and dysphonia after lesioning surgery can be expected to improve over time. The impact of these complications on daily life is limited. On the other hand, visual field impairment and hemiplegia due to cerebral infarction have a greater impact on daily life, and the complications associated with pallidotomy are considered more serious.[7,8] Because the PTT is anterior to the red nucleus and medial to the subthalamic nucleus (STN), it can be captured along the same trajectory as the thalamic vim nucleus when using a parietal lobe approach.

Coenen *et al*. conducted an earlier conceptual study of the parietal approach.[2] They studied the stimulation of the STN and cerebello-thalamic tract from the parietal lobe of two patients with Parkinson's disease and tremors.[2] Additional stimulation of the cerebello-thalamic tract more effectively improved tremors than stimulation of the STN alone.[2] The STN is also a target of the basal ganglia-thalamo-cortical circuit.[2] Coenen *et al*. revealed that a single DBS electrode simultaneously stimulated two circuits.[2] In the present case, coagulation was performed because the patient refused DBS; however, it is theoretically possible to simultaneously stimulate PTT and Vim in treating dystonic tremors by applying an eight-contact DBS electrode in the parietal approach.

Multiple target lesions have been reported for the treatment of dystonia. Hassler *et al*. performed unilateral Forel H1, Voi, and Voa lesioning for cervical dystonia.[9] This method is the same as ours for simultaneous PTT and Vim lesioning. Hassler *et al*. observed motor neglect and hemiplegia in 16% and 2.3% of patients, respectively.[5] Loher *et al*. examined the zona incerta and Vo lesions in 111 patients with cervical dystonia, in which 16% and 15% of patients showed hemiplegia and dysarthria, respectively.[9] Savas *et al*. recently reported the results of RF ablation of the unilateral Forel H field and motor thalamus in 50 patients with dystonia, including 18 with dystonia tremors.[11] The authors' motor thalamus was 3–4 mm posterior, 12–13 mm lateral, and 1 mm superior to the midpoint of the anterior commissure-posterior commissure,[11] suggesting a posterior part of the Vo to the anterior part of the Vim region. Thus, the target of Savas *et al*. was nearly the same as that in our case. They lesioned the Forel H field using a side-outlet electrode and the motor thalamus using a straight electrode. The BFMDRS in patients with dystonic tremors showed a 65.6% improvement, and the evaluation of tremors using the unified parkinson's disease rating scale (UPDRS) tremor score showed an 83.3% improvement.[11] Transient slight hemiparesis on the contralateral side of the surgery was observed in 16% of the patients, which spontaneously resolved in the subsequent weeks.[11] However, the safety of lesioning multiple targets has not been established. As both the pallidal and cerebellar circuits are involved in motor function, excessive lesioning may result in severe hypotonia. Since Hassler *et al*. reported hemiparesis,[4] bilateral combined lesioning may lead to serious complications. Thus, the dual-target approach appears to be optimal for DBS. In addition, the long-term effect of this surgery on dystonic tremors is unknown due to the short follow-up period.

CONCLUSION

The present case suggests that in dystonic tremors involving abnormatilites of the basal ganglia-thalamo-cortical and cerebello-thalamo-cortical circuits, a single electrode can be used to approach both circuits through the parietal lobe approach.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

REFERENCES

1. Buhmann C, Moll CK, Zittel S, Münchau A, Engel AK, Hamel W. Deep brain stimulation of the ventrolateral thalamic base and posterior subthalamic area in dystonic head tremor. Acta Neurochir Suppl 2013;117:67-72.
2. Coenen VA, Rijntjes M, Prokop T, Piroth T, Amtage F, Urbach H, *et al.* One-pass deep brain stimulation of dentato-rubro-thalamic tract and subthalamic nucleus for tremor-dominant or equivalent type Parkinson's disease. Acta Neurochir (Wien) 2016;158:773-81.
3. DeSimone JC, Archer DB, Vaillancourt DE, Wagle Shukla A. Network-level connectivity is a critical feature distinguishing dystonic tremor and essential tremor. Brain 2019;142:1644-59.
4. Hassler R, Dieckmann G. Stereotactic treatment of different kinds of spasmodic torticollis. Confin Neurol 1970;32:135-43.
5. Hassler R. Stereotaxic treatment for spasmodic torticollis. In: Stereotaxy of the human brain. Stuttgart New York: G Thieme; 1982. p. 522-31.
6. Horisawa S, Fukui A, Kohara K, Kawamata T, Taira T. Unilateral pallidotomy in the treatment of cervical dystonia: A retrospective observational study. J Neurosurg 2019;134:216-22.
7. Horisawa S, Fukui A, Takeda N, Kawamata T, Taira T. Safety and efficacy of unilateral and bilateral pallidotomy for primary dystonia. Ann Clin Transl Neurol 2021;8:857-65.
8. Horisawa S, Kohara K, Nonaka T, Fukui A, Mochizuki T, Iijima M, *et al.* Unilateral pallidothalamic tractotomy at Forel's field H1 for cervical dystonia. Ann Clin Transl Neurol 2022;9:478-87.
9. Loher TJ, Pohle T, Krauss JK. Functional stereotactic surgery for treatment of cervical dystonia: Review of the experience from the lesional era. Stereotact Funct Neurosurg 2004;82:1-13.
10. Paoli D, Mills R, Brechany U, Pavese N, Nicholson C. DBS in tremor with dystonia: VIM, GPi or both? A review of the literature and considerations from a single-center experience. J Neurol 2023;270:2217-29.
11. Savas A, Bayatli E, Eroglu U, Akbostanci MC. Combined unilateral radiofrequency lesioning of the motor thalamus, field of Forel, and zona incerta: A series of cases with dystonia. Neurosurgery 2022;90:313-21.
12. Trompette C, Giordana C, Leplus A, Grabli D, Hubsch C, Marsé C, *et al.* Combined thalamic and pallidal deep brain stimulation for dystonic tremor. Parkinsonism Relat Disord 2022;103:29-33.
13. Tsuboi T, Au KL, Deeb W, Almeida L, Foote KD, Okun MS, *et al.* Motor outcomes and adverse effects of deep brain stimulation for dystonic tremor: A systematic review. Parkinsonism Relat Disord 2020;76:32-41.
14. Tsuboi T, Wong JK, Eisinger RS, Okromelidze L, Burns MR, Ramirez-Zamora A, *et al.* Comparative connectivity correlates of dystonic and essential tremor deep brain stimulation. Brain 2021;144:1774-86.
15. Yamahata H, Horisawa S, Hodotsuka K, Kawamata T, Taira T. Long-term successful outcome of dystonic head tremor after bilateral deep brain stimulation of the ventral intermediate and ventro-oral internus nuclei: A case report and literature review of dystonic head tremor. Stereotact Funct Neurosurg 2021;99:107-12.

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