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Case Report

Deep brain stimulation for essential tremor in patients with ventriculomegaly

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ABSTRACT

Background: Deep brain stimulation of the nucleus ventralis intermedius (VIM-DBS) is considered a safe and effective treatment for medically intractable essential tremor (ET). However, ventriculomegaly can provide a surgical challenge, as there is an increased risk of breaching the ventricle during the procedure, with potential risk of intraventricular hemorrhage and target displacement.

Case Description: In this case series, we report successful bilateral VIM-DBS in a 72-year-old and 69-year-old female ET patient with significant ventriculomegaly. VIM-DBS therapy provided an excellent tremor response. After 5 years, a ventriculoperitoneal shunt was implanted in the first patient due to an incomplete Hakim-Adams triad, with significant improvement in gait and cognition.

Conclusion: To the best of our knowledge, we present the first report on VIM-DBS in ET patients with ventriculomegaly and illustrate that VIM-DBS can provide an excellent tremor response in patients with medically intractable ET, even in the context of marked ventriculomegaly.

Keywords: Deep brain stimulation, Essential tremor, Ventriculomegaly

INTRODUCTION

Essential tremor (ET) is the most common movement disorder with a prevalence of 5–6% in the general population. The incidence increases with advancing age, with age peaks in the second and sixth decades of life. Although the natural history typically involves a slow progression of tremor intensity with age, ET is generally characterized by a 4–12 Hz postural and kinetic tremor, mainly affecting the arms and less frequently the head, voice, and lower extremities.[12]

First- and second-line medical treatments tend to improve the tremor in approximately 50% of ET patients.[6] When the best medical treatment does not provide adequate tremor suppression, surgical treatments including thalamotomy and deep brain stimulation (DBS) are considered.[7] The application of stereotactic thalamotomy is limited to unilateral intervention due to the risk of irreversible adverse events after bilateral treatment. Therefore, thalamic DBS of the nucleus ventralis intermedius (VIM-DBS) is considered the most safe and effective treatment for medically refractory ET, although evidence is growing to support the posterior subthalamic area (PSA) as an alternative target.[2,8]

However, ventriculomegaly as in normal pressure hydrocephalus (NPH) can provide a surgical challenge. Ventriculomegaly increases the risk of breaching the ventricle during the procedure, with potential risk of intraventricular hemorrhage and target displacement. Hence, neurosurgeons may refuse to operate on ET patients with ventriculomegaly. In this case series, we report successful bilateral VIM-DBS in ET patients with significant ventriculomegaly and an increased anterior and posterior commissure (AC-PC) line.

CASE REPORT

Case 1

A 72-year-old female presented at the neurology outpatient clinic due to disabling bilateral tremor with onset at the age of 61. Cerebral imaging including a dopamine transporter scan displayed an enlargement of the lateral and third ventricles but was otherwise considered normal. Despite best medical treatment including primidone, propranolol, gabapentin, pregabalin, and clozapine, she experienced an insufficient suppression of the tremor over time. After a multidisciplinary team meeting, she was referred to our center at the age of 76 to discuss VIM-DBS.

At the time of presentation, she suffered a disabling bilateral action tremor of the upper limbs, as well as a resting and postural tremor of both arms. On the Fahn-Tolosa-Marin (FTM) scale, she scored 66 points, indicating the substantial impact on her overall quality of life (QOL). The preoperative diagnostic magnetic resonance imaging (MRI) displayed a supratentorial ventriculomegaly with an Evans index of 0.35 [Figure 1a]. No clinical findings compatible with the Hakim-Adams triad were found at the time. Planning of the VIM-DBS was done using a preoperative 3T MRI and Elements© software (Brainlab AG, Munich, Germany). The AC-PC line measured 28.6 mm and the width of the third ventricle at AC-PC level 14.1 mm [Figure 2a]. After fitting these measurements into the theoretical calculations of the bilateral VIM-targets, the autosegmentation of the software helped us to optimize the trajectories. Because the VIM is more superficially located than the PSA, the VIM was chosen as the target of interest. The coordinates relative to the midcommissural point (MCP) on the left and right side were, respectively, −5/5.43 (x), −17.5/15 (y) and 0/0 (z) mm with a slide of 58.6°/57.8° of the semicircular ring and the ring positioned in 23.5°/48.2°.

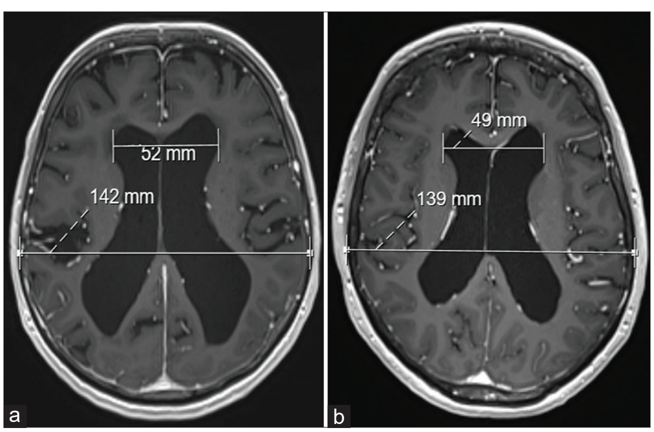

Figure 1: Preoperative axial T1 magnetic resonance imaging demonstrating supratentorial ventriculomegaly in both patients, as indicated by an Evan's index of 0.35 in (a) patient 1 and (b) patient 2.

Surgery was performed under sedation, using macrostimulation to verify the position of the microelectrode. Both directional leads (Vercise Cartesia, DB-2203–30, Boston Scientific) were implanted on the predefined targets at AC-PC level [Figure 3a]. The VIM-DBS therapy provided an excellent tremor response which resulted in an increase of overall QOL.

At the age of 77, she developed a magnetic gait but no decrease of attention span or urinary incontinence. Repeated lumbar punctures according to the Fisher test were performed with improvement. After a multidisciplinary team meeting, she was diagnosed with idiopathic NPH with an incomplete Hakim-Adams triad. A ventriculoperitoneal shunt was implanted using the Keen's point approach [Figure 3a].

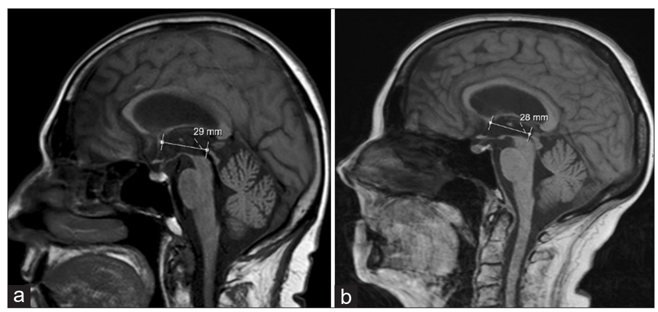

Figure 2: Preoperative sagittal T1 magnetic resonance imaging displaying an increased anterior and posterior commissure line in (a) patient 1 - 28.6 mm and (b) patient 2 - 28.0 mm.

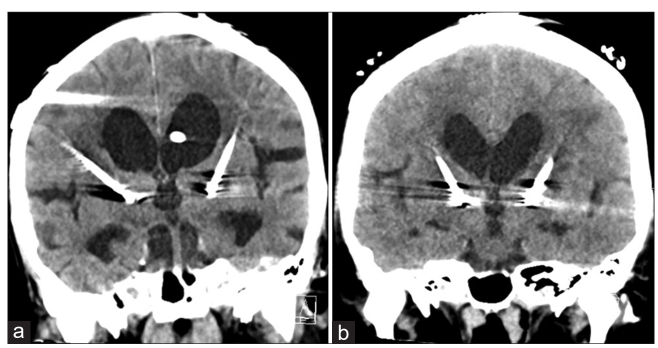

Figure 3: Postoperative coronal computed tomography in (a) patient 1 and (b) patient 2 shows the position of the electrodes at the ventralis intermedius in relation to the ventricles. In (a) patient 1, the presence of a ventriculoperitoneal shunt is additionally demonstrated, implanted using the Keen's point approach.

After the procedure and during a total follow-up period of 6 months, she experienced a significant improvement in gait and cognition as well as an excellent tremor response.

Case 2

A 69-year-old female patient was referred to the neurosurgery outpatient clinic after a multidisciplinary team meeting due to a persisting debilitating tremor despite best medical treatment including primidone, propranolol, and clozapine. Cerebral imaging including a dopamine transporter scan demonstrated an enlargement of the lateral and third ventricles but was otherwise considered normal.

At the time of presentation, she had an action tremor of the upper limbs, as well as a resting head tremor and postural tremor of both arms. She scored 71 points on the FTM scale, signifying the considerable influence on her QOL. The preoperative diagnostic MRI demonstrated a supratentorial ventriculomegaly with an Evans index of 0.35 [Figure 1b]. She had no clinical findings compatible with the Hakim-Adams triad.

Planning of the VIM-DBS was done using a preoperative 3T MRI and Elements© software (Brainlab AG, Munich, Germany). The AC-PC line measured 28.0 mm and the width of the third ventricle at AC-PC level 10.8 mm [Figure 2b]. The autosegmentation of the software helped us to optimize the trajectories after fitting these measurements into the theoretical calculations of the bilateral VIM-targets. Because the VIM is more superficially located than the PSA, the VIM was chosen as the target of interest. The coordinates relative to the MCP on the left and right side were, respectively, −5.53/−4.17 (x), −15.41/14.14 (y) and 0.48/0.02 (z) mm with a slide of 83.8°/88.6° of the semicircular ring and the ring positioned in 28.9°/26°.

Surgery was performed under sedation, using macrostimulation to verify the position of the microelectrode. Both directional leads (Vercise Cartesia, DB-2203-30, Boston Scientific) were implanted on the predefined targets at AC-PC level [Figure 3b]. The VIM-DBS therapy provided an excellent tremor response which resulted in an increase of overall QOL. During a total follow-up period of 3 months, she remained symptom-free. After 1 year, she scored 39 on the FTM scale, revealing the postoperative improvement of her symptoms.

DISCUSSION

Thalamic DBS is deemed the most safe and effective treatment for medically intractable ET, presumably because the circuits involved in the pathophysiology of different tremors all seem to converge at the ventrolateral thalamic area. Multiple structures in this area have been targeted, of which the VIM and the PSA are the most frequently mentioned. The VIM is the classic target for neurosurgical treatment for ET. It is connected to the contralateral deep cerebellar nuclei by the dentatorubrothalamic tract (DRTT) and projects to the ipsilateral motor and association cortices. According to a recent literature review, bilateral VIM-DBS is estimated to provide better tremor control in 66–78% of patients. In a prospective study of 11 tremor patients, *post hoc* tractographic analysis demonstrated higher rates of tremor control when the electrodes were located inside or in proximity with the DRTT.[3] Unfortunately, habituation to stimulation is frequently reported and thalamic DBS is not without potential complications.[4]

The most frequently used stereotactic coordinates for the VIM are based on statistical estimations in relation to the third ventricle and the mid-commissural line, initially derived from the stereotactic atlases of Schaltenbrand G and of Talairach *et al*.[7,9] According to these estimations, the VIM is situated in the posterior fourth of the AC-PC line at AC-PC level and approximately 15 mm from the midline. Because the VIM is located laterally from the third ventricle, the width of the third ventricle should be taken into consideration. When an abnormal width of the third ventricle is suspected, the rule of Tasker can be applied. This implies the calculation of the laterality as 11.5 mm + (width of the third ventricle/2).[1] In our cases, we have used the Elements© software (Brainlab AG, Munich, Germany) as an extra control. The autosegmentation of this software on preoperative 3T MRI and diffusion tensor imaging allowed us to identify relevant anatomical brain structures and helped us to optimize the trajectories.

In 1972, Velasco *et al*. suggested that the PSA may be a valuable alternative to VIM targeting.[11] More recently, it has been evinced that the zona incerta and the prelemniscal radiation, both encompassed by the PSA, are the most effective targets in the PSA.[10] The clinical effect is considered to originate from direct modulation of the DRTT. Although there is increasing evidence in the literature to support the PSA as an alternative target, systematic reviews have not been able to demonstrate superiority of PSA targeting in the long term.[5] Altogether, there is insufficient evidence to conclusively support a substantial disparity in efficacy between VIM- and PSA-DBS. As we have good experiences with traditional VIM targeting and this nucleus is more superficially and laterally located than the PSA, we have opted to use the VIM as a target in our patients.

Thalamic DBS of the VIM or PSA is not without potential complications. In patients without significant macroscopic anatomical abnormalities, electrode insertion can cause intracerebral hemorrhage (0.5–1.5%), wound infection (1.7–5.4%) and hardware-related complications (1.4–3.8%). Furthermore, neurological side effects of bilateral VIM-DBS include ataxia (56–85.7%), dysarthria (22–75%), dysphasia (18.8%), transient hemiparesis (6.7%), and stimulation-induced paresthesia (5.9%).[4]

Ventriculomegaly and an increased AC-PC line can provide an additional surgical challenge, as there is a theoretically increased risk of breaching the ventricle and target displacement. Therefore, neurosurgeons may refuse to perform thalamic DBS on patients with enlarged ventricles. However, as observed in our own practice and depicted in this report, patients with ventriculomegaly should not be denied thalamic DBS in appropriate indications. With careful preoperative planning and fine adjustment of the coordinates to include a more lateral entry, favorable outcomes can be attained.

CONCLUSION

To the best of our knowledge, we present the first report on VIM-DBS in ET patients with ventriculomegaly and illustrate that, with careful preparation; VIM-DBS can provide an excellent tremor response in patients with medically intractable ET, even in the context of marked ventriculomegaly.

Author's contributions

1. Research project: A. Conception, B. Organization, C. Execution; 2. Statistical Analysis: A. Design, B. Execution, C. Review and Critique; 3. Manuscript Preparation: A. Writing of the first draft, B. Review and Critique.

RB: 1C, 2B, 3A; SV: 1B, 2A, 3B; FV: 1C, 2C, 3B; WM: 1B, 2C, 3B; SDF: 1B, 2C, 3B; JVL: 1B, 2C, 3B; DV: 1B, 2C, 3B; OVD: 1A, 2C, 3B;

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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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.

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