INTRODUCTION

Neurosurgical treatment is considered as an effective treatment option for patients with essential tremor (ET), since approximately 50% of the ET population is refractory to medical treatment [1]. Historically, various methods, from ablative procedures to deep brain stimulation (DBS) have been used, each having its own strengths and weaknesses. With the recent development of magnetic resonance guided focused ultrasound (FUS), a new trend in the lesioning procedure for ET has emerged [2]. Although each treatment can be complementary to each other, clinicians inevitably face difficulties in deciding which surgical technique to use or which target needs to be operated on for the second treatment. In recurrent case after lesioning procedure, it is uncertain whether stimulating previously injured neural structures can achieve the same effectiveness to control tremor. Moreover, it is uncomfortable to set the intracranial surgical tract due to thalamic scarring during the DBS. In contrast, lesioning after failed DBS can be very burdensome to ablate nearby areas with electrical devices.

Here, we present a case of recurrent tremor after ablation of the ventral intermediate nucleus of the thalamus (VIM), which was effectively managed with posterior subthalamic area (PSA) DBS, and review previous reports with similar experiences.
CASE REPORT

A 49-year-old right-handed man visited our outpatient clinic with 30-year history of bilateral hand tremor. He was diagnosed with ET and managed by left VIM thalamotomy at another medical center 13 years ago. His tremor improved immediately after the thalamotomy; however, it recurred several years ago and progressively worsened. The patient had no relevant medical history. Initial laboratory findings were normal, including a hormonal work-up. He was already taking propranolol prescribed at another hospital, which did not work. Primidone was additionally prescribed for symptom control, but its use had to be restricted owing to the associated side effects, including severe nausea and dizziness. His tremor was assessed using clinical rating scale for tremor, with an initial score of 11 points for Part A (tremor location and severity rating), 11 points for Part B (functional rating), and 12 points for Part C (functional disabilities) (Fig. 1A). The clinical score improved to 7, 6, 7 points after the addition of baclofen, but symptomatic control failed after about a year. In particular, the tremor handicapped him in performing activities of daily living such as writing, using chopsticks, and drinking water. The patient finally decided to undergo unilateral DBS due to relatively high risks of stimulation-induced side effects of bilateral DBS. Preoperative brain magnetic resonance imaging (MRI) revealed a small cystic scar in the left thalamus, in which the correctly placed VIM lesion was presumed to be sufficiently performed based on Schaltenbrand-Wahren atlas. The target of stimulation was proposed to be the left PSA because there was a possibility of ineffective stimulation to the previous surgical site. The coordinates were 5.7 mm anterior to the posterior commissure, 3.9 mm below the anterior and posterior commissure line, and 11.7 mm lateral to midline (Fig. 2). The trajectory was set anterolateral to the previous thalamic lesion. The surgery was performed under local anesthesia. Intraoperatively, some kinetic cells were identified, but no significant tremor cells were observed during the microelectrode recording. With microstimulation along the electrode trajectory, monopolar stimulation at the ventral contacts (0 contact, PSA) confirmed a more effective tremor control than dorsal contacts (possibly near VIM). Postoperatively, the patient showed significant tremor reduction and functional improvement (3 points for Part A, 3 points for Part B, and 2 points for Part C) without stimulation-induced side effects (Fig. 1B). He continued using unipolar stimulation at contact 0, 1 with the following parameters: 2.7 mA, pulse width 60 μsec, and frequency of 160 Hz. This benefit persisted without any additional medications at 2-year follow-up.

Ethical statements

This study was approved by the Institutional Review Board of Ulsan University Hospital (No. 2022-05-015), and the requirement for the patient’s written consent was waived.

DISCUSSION

In the case of lesioning for ET, the popularity of conventional ra-

![Archimedean spiral and line drawings by the patient presented herein. The drawing on the left was drawn at his first visit to our clinic (A), and that on the right was drawn one day after deep brain stimulation (B).](https://doi.org/10.52662/jksfn.2022.00171)
diofrequency (RF) ablation has declined in the era of DBS, but the recent rise in FUS has regained popularity along with less invasive gamma knife thalamotomy [2]. Nevertheless, the fundamental disadvantage of lesioning is the potential risk of loss of effect when follow-up. Considering previous experiences with RF ablation, successful tremor control by thalamotomy can diminish in up to 15% of cases [3]. Even in FUS thalamotomy, early clinical reports documented that five of the 56 patients experienced recurrent tremor with tremor score worsening by 23.2% within 1 year [2]. Therefore, there is always a demand for complementary treatments for such recurrent cases, including reattempting of lesioning or DBS.

DBS has been adopted by centers worldwide to suppress tremor due to its reversibility and flexibility in treating dominant disease symptoms [4]. A randomized controlled study showed that thalamic stimulation and thalamotomy are equally effective for drug-resistant tremor. However, DBS has fewer adverse effects and greater functional improvement [5].

Although concerns remain that previous lesioning procedure could limit the effectiveness of future DBS, previous studies have shown that DBS can be successfully performed following RF, gamma-knife and FUS thalamotomy [6-10].

Since first introduced by Benabid et al. [11] in 1987, the VIM DBS continues to be used as the traditional target for DBS in the treatment of intractable tremor. Although VIM DBS is still most widely used in managing tremor, it is not the perfect approach due to certain shortcomings, such as the possibility of tolerance, dysarthria, postural instability, or cognitive impairment, which are frequently encountered, especially in bilateral procedures [12]. In addition, since changes in nerve fibers related to tremor cannot be predicted when permanent lesioning is performed in the thalamus, it is difficult to determine whether DBS targeting the vicinity of the preexisting stereotactic lesion is effective. There have been several reports of DBS being successfully performed in the area close
to the previous lesion, but there is no guarantee that it can be applied to all cases, particularly when the intraoperative findings are different from the expected [7-10].

PSA was introduced as an alternative target for ET by Wertheimer et al. [13] in 1960. It involves the zona incerta and prelemniscal radiation, including afferent fibers from the dentato-thalamic tract, which are possibly related to controlling tremor. It is located lateral to the red nucleus, inferior to the ventrolateral thalamus, posteriomedial to the subthalamic nucleus (STN), and anterior to the medial lemniscus [14,15]. Targeting PSA is relatively easy because nearby structures, such as the STN and red nucleus, can be seen on MRI and can be used as anatomical landmarks for target setting when compared to the indirect targeting of VIM based on its relationship to the anterior and posterior commissure [16]. Previous studies have shown positive outcomes of PSA DBS for ET management. Murata et al. [14] reported a significant reduction in the tremor scale in 81% of patients after unilateral PSA DBS. Blomstedt et al. [16] also reported that the total score of the tremor was reduced to 60% from baseline to 1-year follow-up; in particular, 95% improvement was seen in the tremor of the upper extremity, accompanied by 66% improvement as anatomical landmarks for daily living. When comparing VIM and PSA as optimal targets for tremor, previous studies have shown that they are not superior in reducing tremor [17]. However, a randomized, double-blind, crossover trial by Barbe et al. [18] identified that PSA DBS is equally effective and more efficient than VIM DBS after 1-year-use. A recent meta-analysis also demonstrated modest superior efficacy of PSA stimulation for treating refractory ET in the short-term follow-up [19]. In addition, simultaneous targeting of VIM and PSA via one trajectory is possible because simply advancing the electrode more deeply from VIM can reach the PSA accurately [20]. This can compare the effectiveness between PSA and VIM or prepare one contingency plan in choosing the stimulation site for individualized adjustment according to the patient’s symptoms.

CONCLUSION

We report the successful treatment of recurrent ET using PSA DBS after VIM thalamotomy. Although there are various surgical treatment options for recurrent ET, DBS targeting PSA can provide a favorable solution to compensate for the unsatisfactory outcomes from VIM thalamotomy.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

AUTHORS’ CONTRIBUTION

Seung Un Lee: Writing - Original draft, Formal analysis, Data curation
Na Young Jung: Conceptualization, Writing - review & editing, Supervision

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