

POSTEROVENTRAL PALLIDOTOMY IN MEDICALLY INTRACTABLE POSTAPOPLECTIC MONOCHOREA: CASE REPORT

Seung-Jin Choi, M.D.,* Sang-Won Lee, M.D.,* Moon-Chan Kim, M.D.,* Jeong-Yi Kwon, M.D.,† Choon-Keun Park, M.D.,* Jae-Hoon Sung, M.D.,* Jae-Taek Hong, M.D.,* and Hee-Kyung Woo, M.D.*

**Department of Neurosurgery and †Rehabilitation Medicine, St. Vincent Hospital, College of Medicine, The Catholic University of Korea, Seoul, Korea*

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BACKGROUND

Posteroventral pallidotomy is a widely accepted surgical procedure for treating medically intractable Parkinson's disease and Levo-dopa induced dyskinesia. In the surgical treatment of hyperkinetic movement disorders, generalized dystonia has recently become a favorable indication of posteroventral pallidotomy. However, a commonly recognized surgical procedure for treating choreiform movement disorders has not yet been established. Here we present an unusual experience of a posteroventral pallidotomy performed to treat a medically intractable monochorea caused by a vascular insult on the basal ganglia.

CASE PRESENTATION

A 63-year-old female presented with choreiform movement of the left upper limb that she had suffered for 5 months. She was found to have a hemorrhagic infarction in the right putaminal area. No other abnormal lesions were shown by magnetic resonance imaging except for a widening of the right cerebellopontine cistern because of an acoustic neurinoma removed 5 years previously. Despite medication with a dopamine antagonist, choreiform movement of the left limb had not improved, and the patient complained of rigidity and slowness of ambulation owing to the side effects of the medicine. A right posteroventral pallidotomy was performed with macrostimulation for a physiologic confirmation of the globus pallidus internus (GPi), which is the conventional target for Parkinson's disease. After coagulating the GPi target, the choreiform movement of the contralateral upper limb was completely abolished.

The postoperative course was uneventful and no recurrence of chorea was observed over a follow-up period of 6 months.

CONCLUSION

Stereotactic surgery for hyperkinetic movement disorders is not as common a procedure as that used for treating Parkinson's disease. Furthermore, there have been few reports of pallidal surgery for treating the chorea caused by an ischemic insult. However, on the basis of the current concept that varying types of hyperkinetic disorders may have a common pathophysiological mechanism, a posteroventral pallidotomy may be an alternative surgical procedure for treating medically intractable postapoplectic chorea like in an occasion of dystonia. © 2003 Elsevier Inc. All rights reserved.

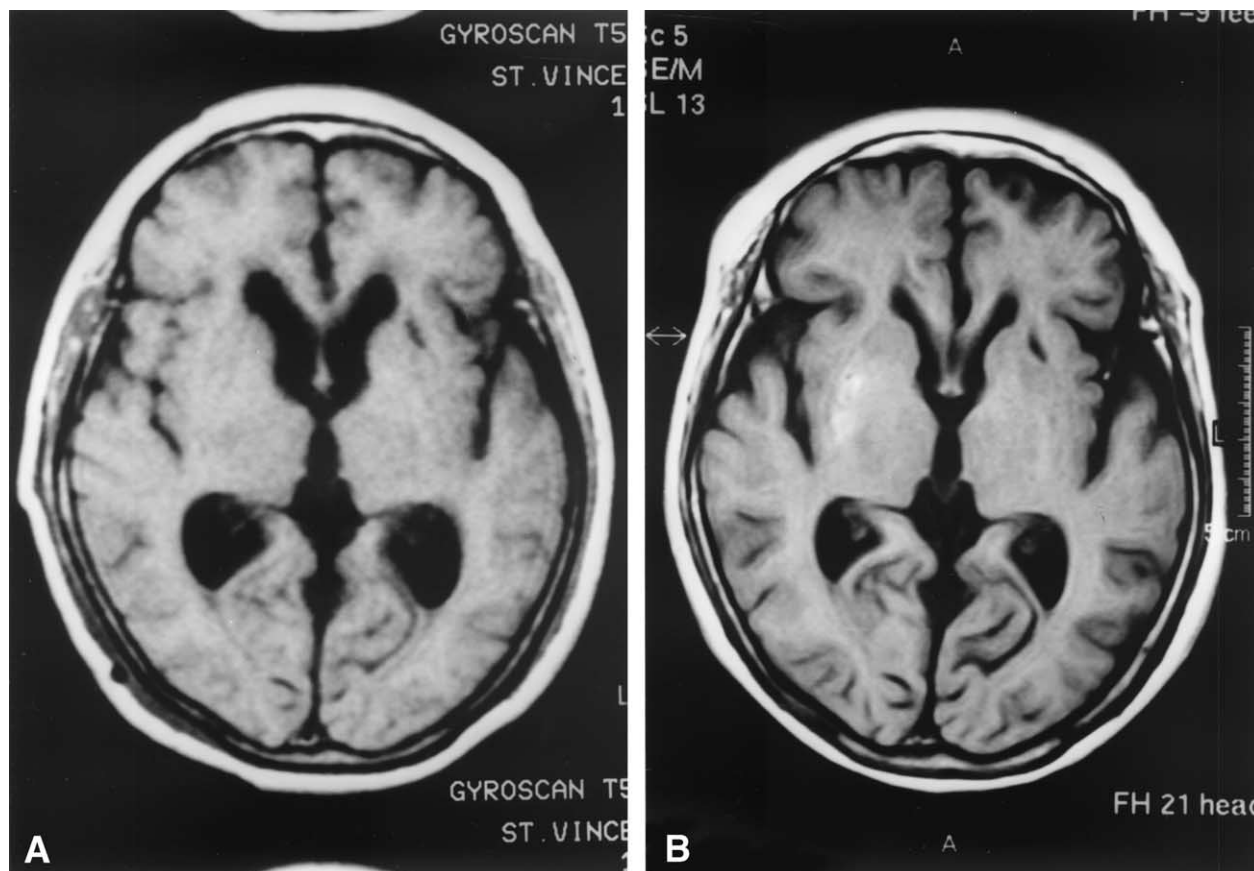
KEY WORDS

Macrostimulation, posteroventral pallidotomy, postapoplectic monochorea, striatal hemorrhagic infarction.

Various stereotactic surgical procedures for treating hyperkinetic movement disorders have been attempted since the first trial in Huntington's chorea by Spiegel and Wycis in 1946 [6]. However, unlike Parkinson's disease, confirmed targets in the brain and surgical methods have not been established as common procedures for treating hyperkinetic disorders. In dystonia, based on the improvement in dystonic symptoms in surgically treated Parkinson's disease, stereotactic surgery is not uncommon. Classically, the thalamus was the main target [1,4] and recently, pallidotomy is becoming a more favorable procedure particularly for primary dystonia [9,12]. However, in choreiform movement disorders as well as in hemiballism, stereotactic surgical treatments have rarely been attempted. Therefore, there is much controversy re-

Address reprint requests to: Dr. Seung-Jin Choi, Department of Neurosurgery, St. Vincent Hospital, 93-6 Chi-dong Paldal-ku, Suwon 442-723 Korea.

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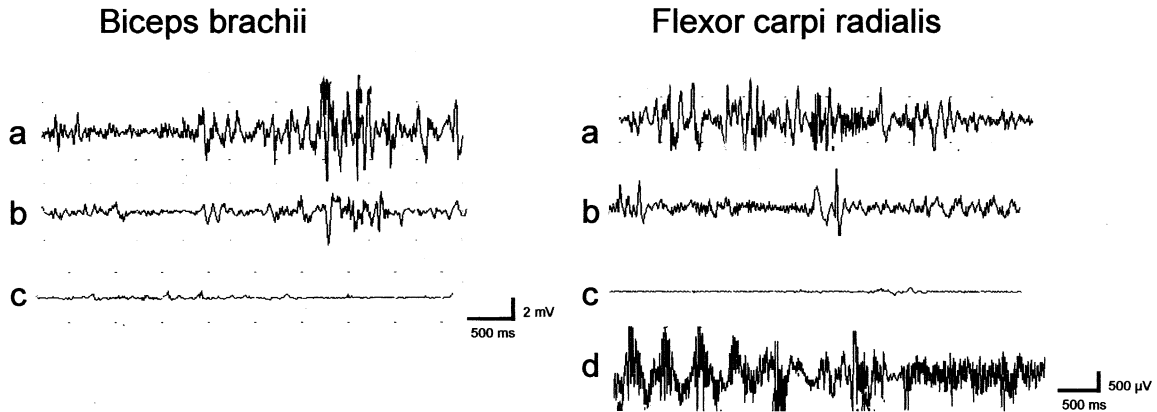
1 T1-weighted MRI scans without gadolinium enhancement performed 5 years before developing monochorea (A), and taken 3 months after the onset of choreiform movement (B) demonstrate a newly developed hemorrhagic infarction in the right putaminal area, which caused the involuntary choreiform movement of the left upper limb.

garding the optimal surgical target in the brain. Here, we present a patient with medically intractable monochorea caused by a hemorrhagic infarction in the putaminal area, who was treated successfully by posteroventral pallidotomy, which is one of the conventional surgical methods for Parkinson's disease.

CASE REPORTS

A 63-year-old female presented with complaints of spontaneous rapid involuntary movement of the left arm and hand for 5 months. Five years before developing monochorea, she had undergone a total tumor removal of right acoustic neurinoma via a retromastoid craniotomy. Magnetic resonance imaging (MRI) that had been taken postoperatively, revealed no abnormality in both basal ganglia (Figure 1A). However, a MR examination taken 3 months after the onset of choreiform movement demonstrated a hemorrhagic infarction in the right putaminal area (Figure 1B). The other medical his-

tory of the patient was diabetes mellitus and bronchial asthma. The patient was initially treated with a dopamine antagonist (1.5 mg of haloperidol daily). Even though the haloperidol dose was increased to 6 mg daily for 3 weeks, the choreiform movement of the left upper limb was not diminished and the patient showed some rigidity and slowness of ambulation. Consequently, medication was withheld. We decided on surgical treatment for this medically intractable monochorea, and right posteroventral pallidotomy was performed using macrostimulation for the physiologic confirmation of the pallidal target. The anatomic localization of the target was performed with MRI, which was selected 1.5 mm anterior to the midpoint of the intercommissural line, 20 mm lateral toward the right side from the midline, and 4 mm inferior to the intercommissural plane. Stimulation was carried out using a macro-electrode (diameter 1.1 mm, exposed tip length 3 mm) to confirm the sensorimotor area of the right globus pallidus internus (GPi), which is the conventional target of the pallidotomy

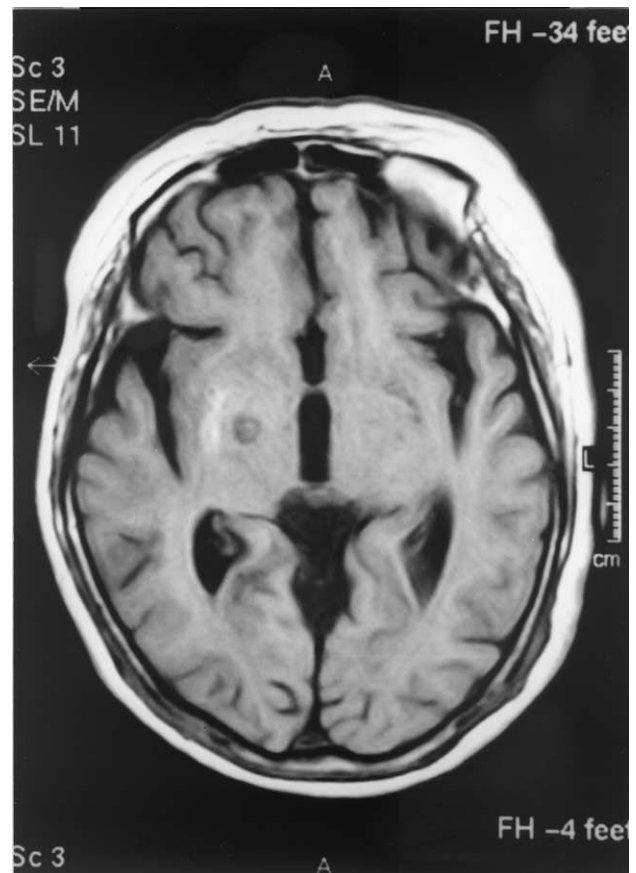


2 Intraoperative EMG activity of the left biceps brachii and flexor carpi radialis was recorded by surface electrodes [a Viking IV (Nicolet, UK) electromyography was used]. Before the coagulation of the right GPi, the dysrhythmic involuntary motor unit potentials at rest position (chorea discharges) were noticed in the biceps brachii and the flexor carpi radialis. During coagulation, these activities were significantly lower and completely abolished immediately after coagulation. However, the motor unit potentials on violation were not impaired. (a) Involuntary dysrhythmic motor unit potentials (MUPs) at the rest position before lesion making. (b) Marked decrease in the MUPs at the rest position during lesion making. (c) Abolishment of MUPs at the rest position after lesion making. (d) Preservation of normal MUPs on violation in flexor carpi radialis after lesion making.

for Parkinson's disease. The desired sensorimotor area of the GPi was decided by stimulation with square wave pulses at a rate of one per second, a frequency of 50 Hz and a voltage threshold of 1.3 volts to elicit the capsular sign (contralateral tonic limb contraction) [10]. The intraoperative visual evoked potential was monitored to distinguish and avoid visual complication. A reversible radiofrequency lesion first was produced at 45°C for 30 seconds to detect any neurologic side effects. After a permanent lesion was produced at 80°C for 60 seconds, the choreiform movement of the left arm and hand nearly completely disappeared. The monitoring of the intraoperative EMG also showed the disappearance of repetitive reciprocal discharges of the biceps brachii and the flexor carpi radialis muscles in the resting position of the left upper limb (Figure 2). After making a second lesion just 2 mm above the first lesion along the track, the patient's limb was closely observed for several minutes and no recurrence of choreiform movement was confirmed. The postoperative course was uneventful without a recurrence of choreiform movements over a follow-up period of 6 months at present. The postoperative MRI is illustrated in Figure 3.

DISCUSSION

Lesions of the basal ganglia caused by a vascular insult commonly induce involuntary hyperkinetic



3 Postoperative T1-weighted MRI scan without intravenous enhancement demonstrates an actual posteroventral pallidotomy lesion shown in the area of the right globus pallidus internus.

movements, and dystonia is the main pattern when movement disorder takes place [2]. Choreiform movement is an uncommon pattern of dyskinesia if the lesions are involved in the lentiform nuclei, particularly in the putamen, which are more likely to cause dystonia. As in this case with choreiform movement caused by a hemorrhagic infarction in the contralateral putaminal area, it appears that there is no absolutely confined anatomic lesion of basal ganglia that manifests only one specific pattern of abnormal movement. Traditionally, hyperkinetic movement disorders are classified on the basis of their clinical appearance, but the differences in the pathophysiological mechanisms between the types of hyperkinesia are uncertain. In MPTP-treated primate models of parkinsonism, a significant increase in tonic neural firings has been observed in the globus pallidus internus (GPi) and the subthalamic nucleus (STN) [13]. In addition, increased neural discharge of the GPi and STN also has been disclosed in parkinsonian patients [14,18]. This is because a loss of dopamine in the striatum results in a disinhibition of the GPi via the direct and indirect pathways. In the indirect pathway, a neuronal discharge of the STN is increased as a result of a diminished or loss of inhibitory input transmitted from the globus pallidus externus (GPe), which leads to an increase in the neuronal discharge of the GPi. The abnormally increased inhibitory output from the GPi induces a reduction in the activity of the thalamocortical circuit. This results in the motor disturbances associated with parkinsonism. In hyperkinetic movement disorders, the pathophysiological mechanisms have not been well established compared to Parkinson's disease. Classically, the subthalamic nucleus is well known as the anatomic location of the lesion in hemiballismus. Animal experiments of a subthalamic lesion produce ballistic movement [3,7]. However, the loss or inactivation of the STN control on the globus pallidus internus is proposed as an important factor that induces various patterns of hyperkinetic disorders as well as ballistic movement [5]. Suarez et al reported a case of hemiballismus caused by a subthalamic hemorrhage in a patient who underwent a microelectrode recording (MER) of the GPi during pallidotomy [16]. They revealed that the neuronal activity of the GPi was characterized by unpatterned low-frequency neuronal discharge, which suggests the loss or diminution of subthalamic influences to the GPi. Vitek et al reported that in generalized dystonic patients, low neuronal activities with an irregular pattern were also observed in the sensorimotor area of the GPi, similar to a patient showing hemiballismus [17]. In addition to the

low neuronal firing rates, they suggested that differences in the pattern of discharge, the degree of synchronization and the altered responsiveness of the pallidal neurons to a sensory input are important factors causing various clinical types of hyperkinesias. Hashimoto et al recently reported altered neuronal firing patterns with a low frequent discharge in a chorea patient who underwent a MER of the GPi [8]. This suggests that chorea can also be induced by an alteration of the normal pallidal neuronal activity as in dystonia and hemiballismus. Posteroventral pallidotomy has become the preferring surgical procedure for generalized dystonia with favorable results frequently being reported [11,12,15]. The ablation of abnormal neuronal activity of GPi by pallidotomy may result in an improvement in chorea by interrupting the unregulated neural circuits as found in dystonia and Parkinson's disease [5].

CONCLUSION

Even though the number of experiences of pallidotomy treating intractable chorea are insufficient for it to be accepted as a significant surgical procedure, it should be considered as alternative surgical treatment if the choreiform movement cannot be controlled by medical therapy.

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COMMENTARY

Choi et al present a case with hemichorea, which was treated with pallidotomy. In fact, pallidotomy has been performed in cases with chorea with varying results. Spiegel and Wysis, and Hassler and Narabayashi, reported several cases with chorea that were treated with pallidotomy. As such, this case cannot be regarded as the first case. However, the authors have a right to say that this procedure is rarely performed in cases with chorea, and in their case, pallidotomy was successful. This case report carries the value of calling to the attention of neurosurgeons the use of pallidotomy in cases with chorea.

Yucel Kanpolat, M.D.

*Department of Neurosurgery
Ankara University School of Medicine
Ankara, Turkey*

Problems are only opportunities in work clothes.”

—HENRY KAISER