Successful Management of Hemorrhage-Associated Hemiballism After Subthalamic Nucleus Deep Brain Stimulation with Pallidal Stimulation: a Case Report

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Key words
- Deep brain stimulation
- Delayed hemorrhage
- Hemiballism: Pallidal stimulation
- Subthalamic nucleus

Abbreviations and Acronyms
CT: Computed tomography
DBS: Deep brain stimulation
MRI: Magnetic resonance imaging
MSA: Multiple system atrophy
STN: Subthalamic nucleus

INTRODUCTION
Deep brain stimulation (DBS) is widely used as an adjunctive treatment for movement disorders such as essential tremor, Parkinson disease, and dystonia, as well as other neurologic and psychiatric diseases. More than 100,000 Medtronic systems have been implanted worldwide in the past 25 years (13). Although the benefits of DBS are well documented, serious complications such as intracranial hemorrhage can occur. Hemorrhage rates for microelectrode-guided stereotactic procedures for movement disorders can range from 0% to 9.5%, as demonstrated by several large studies (1). Certain variables have been associated with increased risk for intracranial hemorrhage with DBS placement. These include the number of passes made, trajectories passing through sulci or near the ependymal surface of the lateral ventricles, the use of microelectrode recording, the use of anticoagulant or antiplatelet medication, as well as hypertension (7).

However, delayed occurrence of intracranial hemorrhage after DBS surgery is rare (18). We present a case of delayed hemorrhage in the subthalamic nucleus after stereotactic implantation of DBS electrodes, which resulted in hemiballism. The hemiballism was then subsequently treated with pallidial DBS.

CLINICAL PRESENTATION
A 54-year-old right-handed male presented with a 13-year history of partially levodopa-responsive Parkinsonism, left-sided bradykinesia, and dystonic cramping worse on the left side than right. He had developed unpredictable motor fluctuations and levodopa-induced dyskinesias, which were not amenable to medical therapy. Preoperatively, his Unified Parkinson Disease Rating Scale (UPDRS)—Part 3/total scores were 33/108 off medication and 15/108 on medication approximately 6 months after he presented to the clinic. His preoperative on-off motor diary is displayed in Table 1. The patient was receiving 81 mg of aspirin daily.

The patient opted to proceed with DBS. Aspirin was discontinued 10 days before the date of surgery, and he underwent stereotactic implantation of bilateral subthalamic nucleus DBS electrodes (Model 3387; Medtronic, Minneapolis, Minnesota, USA). Three microelectrode passes were made on the left side, and 1 microelectrode pass was made on the right side. Surgery was uneventful, and no major fluctuations of blood pressure occurred. Postoperative computed tomography (CT) and magnetic resonance imaging (MRI) showed appropriate placement of the electrodes at the intended targets, and there was no evidence of hemorrhage (Figure 1A). His electrodes were internalized to Activa PC dual-channel internal pulse generator 4 weeks later. Initiation of stimulation resulted in substantial reduction of bradykinesia,
He developed hemiballism involving the right arm and leg. Head CT and MRI of the brain revealed a left-sided, 4-mm peri-electrode hematoma, as well as withdrawal of right electrode (Figure 1B and C). His dopaminergic medications were discontinued, and haloperidol was started in an unsuccessful attempt to reduce the movement disorder. Large-field, low-frequency (c=+0.1-2.3, 20 Hz, 360 PW, 2.5V) stimulation via the left electrode reduced the hemiballism only slightly. The patient failed to gain adequate relief from hemiballism in a 2-week span, and we elected to remove the left STN DBS electrode and place a left GPi DBS electrode using microelectrode recording. His right STN electrode was also advanced back to its original position. Two microelectrode passes were made for left GPi DBS electrode placement. Postoperative CT scan and MRI showed adequate positioning of the electrodes (Figure 1D). Stimulation via the left GPi electrode (c=+1-, 160 Hz, 90 PW, 2.0V) substantially reduced right-sided hemiballism, and fine motor control was improved. However, over several weeks following the second operative procedure, the patient’s clinical condition deteriorated. While the hemiballism remained under good control, he gradually developed symptoms that raised the suspicion for multiple system atrophy (MSA). Follow-up MRI of the brain and re-evaluation of prior MRIs revealed the gradual evolution of cerebellum and brainstem atrophy in a pattern suggestive of MSA (Figure 1E and F).

DISCUSSION

Although hemorrhage is a well-recognized complication of microelectrode-guided DBS surgery, a recent review of the literature revealed only a handful of reports of delayed hemorrhage (3, 18). These reported events occurred within a range of 36 hours to 9 months after DBS electrode implantation with initially negative postoperative imaging. A majority of these patients were not on any active antiplatelet therapy and did not suffer from hypertension. One author, however, reported a delayed, recurrent, and spontaneous hemorrhage after DBS in a patient who was on a Factor Xa inhibitor (14). In our case the hemorrhage is likely from the patient’s fall, which was of sufficient force to dislodge the electrodes from their original site, and aided by the ongoing antiplatelet therapy. This case demonstrates that conventional antiplatelet therapy confers a risk for a potentially disabling movement disorder and stresses the importance of developing evidence-based guidelines to assist neurosurgeons in this matter.

As has been shown previously, high-frequency GPi stimulation can be quite effective in the treatment of hyperkinetic movement disorders such as hemiballism (2, 4, 5, 9, 10, 17). A recent prospective, open-label study showed that GPi DBS is a useful adjunct in treating patients with Huntington disease with prominent chorea that is pharmacoresistant (6). Pallidotomy and thalamotomy have been used successfully for postsubbthalamicotomy hemiballism (11, 15) but seem to offer only modest palliative functional improvement in medically refractory cases. Our case is the third that we know of where high-frequency pallidal stimulation was shown to be effective in treating hemiballism arising after STN DBS (8, 12).

Table 1. Preoperative Medication Motor Diary

<table>
<thead>
<tr>
<th>Day 1</th>
<th>Day 2</th>
<th>Day 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Asleep</td>
<td>4:30</td>
<td>4:00</td>
</tr>
<tr>
<td>Off medication</td>
<td>11:00</td>
<td>6:30</td>
</tr>
<tr>
<td>‘On medication’ without dyskinesias</td>
<td>6:00</td>
<td>2:30</td>
</tr>
<tr>
<td>‘On medication’ without troublesome dyskinesias</td>
<td>2:30</td>
<td>11:00</td>
</tr>
<tr>
<td>‘On medication’ with troublesome dyskinesias</td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

Figure 1. (A) Postoperative, noncontrast, T1-weighted magnetic resonance imaging (MRI) showing adequate placement of bilateral subthalamic nucleus (STN) electrodes with no evidence of hemorrhage. (B) Nonenhanced, axial computed tomography scan obtained after the patient fell, demonstrating peri-electrode hematoma on the left and withdrawal of the electrode on the right. (C) Noncontrast, T1-weighted MRI showing a small hematoma in the left STN and withdrawal of the right electrode. (D) Postoperative, noncontrast, T1-weighted MRI showing replacement of the right electrode in the STN. Also seen is the new left GPi electrode. (E) Noncontrast, T1-weighted MRI showing significant cerebellar atrophy. (F) Noncontrast, T2-weighted MRI showing cruciform T2 hyperintensity within the pons, also known as the hot cross bun sign.
Another unusual aspect of our case report is the extremely slow progression of the patient's condition with delayed manifestation of atypical Parkinsonism occurring over a decade after the initial symptoms. MSA, although rare, can masquerade as idiopathic Parkinson disease. Cases have been reported where incorrectly diagnosed, levodopa-responsive idiopathic Parkinson disease showed positive initial responses to DBS surgery followed by a rapid decline secondary to progression of MSA and subsequent death or severe disability (16).

CONCLUSION
We present a rare case of delayed intracranial hemorrhage in a patient who had undergone placement of bilateral STN-DBS electrodes for medically refractory Parkinsonism. The critical location of the hematoma in the STN caused contralateral hemiballism which was treated successfully by high-frequency stimulation of contralateral GPI. This report emphasizes the need for anticoagulation guidelines for patients with movement disorders and reinforces the role of STN as a potential target for hyperkinetic movement disorders.

REFERENCES

Conflict of interest statement: The authors declare that the article content was composed in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest

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