Deep Brain Stimulation in a Patient with Parkinson’s Disease and Cortical Superficial Siderosis

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Abstract:
Cortical superficial siderosis (cSS) is a rare condition that is regarded as a potential magnetic resonance marker of cerebral amyloid angiopathy (CAA). We describe the case of a 68-year-old man with cSS and Parkinson’s disease (PD) who subsequently exhibited incidental microhemorrhages, which were only detected on magnetic resonance imaging (MRI), at one week after deep brain stimulation (DBS) surgery. cSS is now considered to be a significant risk factor for CAA and future bleeding. Therefore, because DBS surgery is invasive and may increase the risk of intracerebral hemorrhage, the procedure should be performed carefully when managing patients with PD and CAA.

Key words: intracerebral hemorrhage, cortical superficial siderosis, cerebral amyloid angiopathy, Parkinson’s disease, deep brain stimulation

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Introduction
Cortical superficial siderosis (cSS) is characterized by hemosiderin subpial deposits in the cortical sulci over the convexities of the cerebral hemispheres (1) and has been described as a potential magnetic resonance marker of cerebral amyloid angiopathy (CAA) (2). The condition may be frequently accompanied by cerebral microbleeds, and a history of intradural surgery or trauma is a common risk factor for cSS (3). In the general population, the reported prevalence of cSS is low (0.43%) (4). We herein describe the case of a patient with Parkinson’s disease (PD) who underwent deep brain stimulation (DBS) and who subsequently exhibited hemorrhagic changes on magnetic resonance imaging (MRI).

Case Report
A 68-year-old right-handed man with an 8-year history of PD was evaluated due to worsening of the wearing-off phenomenon. He was hospitalized to determine whether DBS was indicated. He had no history of recent falls or hypertension. His medications included levodopa/decarboxylase inhibitor (450 mg/day), entacapone (300 mg/day), rasagiline mesylate (1 mg/day), and rotigotine patch (8 mg/24 h). MRI with fluid-attenuated inversion recovery (FLAIR) showed no sign of significant abnormalities or white matter lesions (Fig. 1A); however, susceptibility-weighted imaging (SWI) detected cSS in the occipital fissure (Fig. 1B). The β-amyloid and phosphorylated tau concentrations were not remarkably elevated. Because the severity of motor fluctuation had increased and the Movement Disorder Society Unified Parkinson’s Disease Rating Scale (MDS-UPDRS) Part III off-state score was 30 at that time, the decision was made to perform DBS.

The implantation of bilateral pallidal electrodes was performed as we previously reported (5). Postoperatively, the patient’s medications were modified, and stimulation tuning
Figure 1. Dynamic changes on imaging before DBS, in the immediate postoperative period, and at the 3-month follow-up examination. (A) T2-FLAIR magnetic resonance imaging before DBS. (B) cSS (arrowhead) was detected on the occipital fissure on SWI just before DBS surgery. (C) Computed tomography in the immediate postoperative period shows the implanted DBS leads (arrow) in the globus pallidus, with no sign of bleeding. (D) Edematous changes (arrowhead) were detected on the left frontal lobe on T2-FLAIR in the immediate postoperative period. (E) Incidental microhemorrhages (arrow) were found on the left frontal lobe on SWI one week after DBS. (F) At the 3-month follow-up examination, edematous changes disappeared on T2-FLAIR. (G) At the 3-month follow-up examination, incidental bleeding (arrow) improved on SWI (G). cSS: cortical superficial siderosis, DBS: deep brain stimulation, FLAIR: fluid-attenuated inversion recovery, MRI: magnetic resonance imaging, SWI: susceptibility-weighted imaging

was performed to improve the motor function. On postoperative Day 1, computed tomography showed no sign of bleeding (Fig. 1C). However, at 1-week follow-up, MRI detected a high FLAIR signal on the left frontal lobe and low-intensity areas were visualized on SWI; these areas were distant from the DBS leads (Fig. 1D, E). At the 3-month follow-up examination, his off times had improved. The severity of the motor function had improved, and the MDS-UPDRS Part III off-state score was 16 points. In addition, the high FLAIR signal lesion and low-intensity areas on SWI had almost disappeared (Fig. 1F, G). The time course of the symptoms and interventions is shown in Fig. 2.

Discussion

In the present case, the PD patient with cSS had incidental microhemorrhages that were only detected on MRI one week after DBS surgery. According to the Modified Boston Criteria, this patient met the criteria for possible CAA because of the presence of cSS on preoperative SWI (6, 7). The presence of cSS has been reported as the most important prognostic factor for the future bleeding risk in patients with CAA (8), which may explain why this case patient had bleeding diathesis.

It is important to note that the bleeding lesion on the left frontal lobe was detected on SWI, but not on computed tomography. There are two explanations. First, the DBS lead placement may have caused microvascular damage. If so, then the bleeding should have been detected on computed tomography. Second, the mechanical stimulation of DBS surgery may have indirectly induced microhemorrhages. In an experimental animal model, microhemorrhages transiently reduced visibility during the acute phase of trauma (9). Hypertension, surgical modality, and male sex were identified as significant factors contributing to hemorrhage after DBS (10, 11). In addition, the microhemorrhages in the present case disappeared on SWI at the 3-month follow-up examination after DBS. A prospective study revealed that microbleeds may continue for as long as 9 years, although they may seem to intermittently disappear and reappear (12), a characteristic that may describe the dynamic brain imaging changes in this case. However, the mechanism remains unknown. Overall, although major complications were not observed in this case, physicians should be aware of the cSS is associated with a risk of hemorrhage when performing DBS. Further studies are required to understand the detailed risk
of bleeding in patients after DBS.

In conclusion, this is one of the initial reports of a patient with PD and CAA who had an atypical bleeding complication following DBS. cSS is now considered to be a significant risk factor for CAA and future bleeding. Therefore, because DBS surgery is invasive and may increase the risk of intracerebral hemorrhage, the procedure should be performed carefully in patients with PD and CAA.

The authors state that they have no Conflict of Interest (COI).

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