

A Rare Patient with STN-DBS Presenting with Delayed-onset and Persistent Peri-lead Edema

Dear Editor,

A 69-year-old male patient with Parkinson's disease (PD) presented due to blurred vision and left ptosis. The patient had received the diagnosis of PD 20 years ago due to left-sided slowness and resting tremor. Thirteen months before, Subthalamic Nucleus Deep Brain Stimulation (STN-DBS) (Medtronic, Minneapolis, MN, USA) was inserted due to the advanced stage of the disease. The surgery had been performed under local anesthesia, and intraoperative microelectrode recording and intraoperative test stimulation had been performed to verify the targets. Activation of the DBS* had provided substantial improvement in the MDS-UPDRS-III scores (65 to 35 points) and enabled the reduction of levodopa equivalent dose by 55%. The follow-up postoperative cranial magnetic resonance imaging (MRI) was normal without signs of edema [Figure 1a]. However, a cranial MRI, which was repeated 1 month after surgery due to complaints of blurry vision, showed left peri-lead edema (PLE) [Figure 1b]. The complaints were mild, and the neuro-ophthalmology examination was evaluated as normal. A 1-week course of dexamethasone 4×1 mg was prescribed, which provided complete resolution of the complaints. However, complaints of double vision and concomitant left ptosis appeared again 8 months later, which had progressed gradually within the following 3-month interval. The neurological exam revealed left third-nerve palsy. Other exams were within normal limits. The newly performed cranial MRI (12 months after surgery) showed progression in the left PLE [Figure 1c]. No signs

of infection were detected (vital signs, hemogram, acute reactant proteins, and lumbar puncture). Taken together, the lesion was considered as delayed PLE, and intravenous dexamethasone 4×4 mg was initiated with a decrement schema over the 7 days. The follow-up MRI performed 2 weeks later showed significant resolution of the edema and contrast enhancement [Figure 1d]. Complaints of double vision also improved after the therapy. At the latest follow-up 3 months later (15 months after surgery), there were no symptoms related to PLE.

In our patient, the PLE was initially observed in the first month after the surgery. The symptoms were resolved with dexamethasone therapy. However, the cranial MRI, performed 12 months after the surgery, showed persistence and progression in the PLE. A 1-week course of dexamethasone provided cessation of the symptoms and marked resolution of the PLE. We believe that this rare PLE, which was at delayed onset and persistent, is worthy of reporting.

Classically, PLE is considered a rare and symptomatic complication of DBS.^[1,2] However, contrary to classical knowledge, recent reports have shown that PLE is a more common and underreported condition.^[3,4] In addition, available evidence suggests that the majority of patients with PLE are asymptomatic.^[4] In the prospective study by Whiting *et al.*,^[4] the follow-up of the 191 leads that were placed in 102 subjects revealed PLE in 15 patients (14.7%). Remarkably, seven patients (6.9%) presented with symptoms related to PLE,

whereas the remaining patients were asymptomatic.^[4] Nazzaro *et al.*^[5] retrospectively investigated the data of 189 patients who underwent 363 STN lead implantations, and they found that non-infectious, non-hemorrhagic, delayed, symptomatic PLE occurred in approximately 3% of implanted leads and was more common in re-implantations (9%) compared to new implantations. Lu *et al.*^[6] reported a rare patient undergoing STN-DBS; Computed tomography imaging performed at the second-month postoperative follow-up revealed hypodensities or high signals surrounding the right lead. Generally, the time of the clinical presentation of PLE is reported to range from days to weeks following DBS surgery^[3] However, a recent paper including a literature review has shown that PLE might appear strictly at the later period of the surgery.^[7] Among the 12 patients included in that study, the average symptom onset time was 84.5 days

postoperatively.^[7] Among these patients, two PD subjects with STN-DBS had presented with PLE at the 305th and 396th days of surgery.^[7] In both of them, the resolution of symptoms was achieved with treatments including steroids, antibiotic therapy, and turning the stimulation off.^[7] In another case report by Ramosa *et al.*,^[3] the authors reported a PD subject undergoing STN-DBS who developed PLE more than 3 months after the surgery.^[3] In this patient, a 3-week course of dexamethasone provided a marked improvement in the PLE.^[3] The interesting aspect of our patient was the delayed onset time of the PLE and its persistence at 12 months after the surgery, which is very rarely reported. In addition, despite the MRIs showing apparent PLE, the clinical manifestations were slightly drawing attention to the importance of the recognition of this major complication, which can be easily overlooked in clinical grounds.

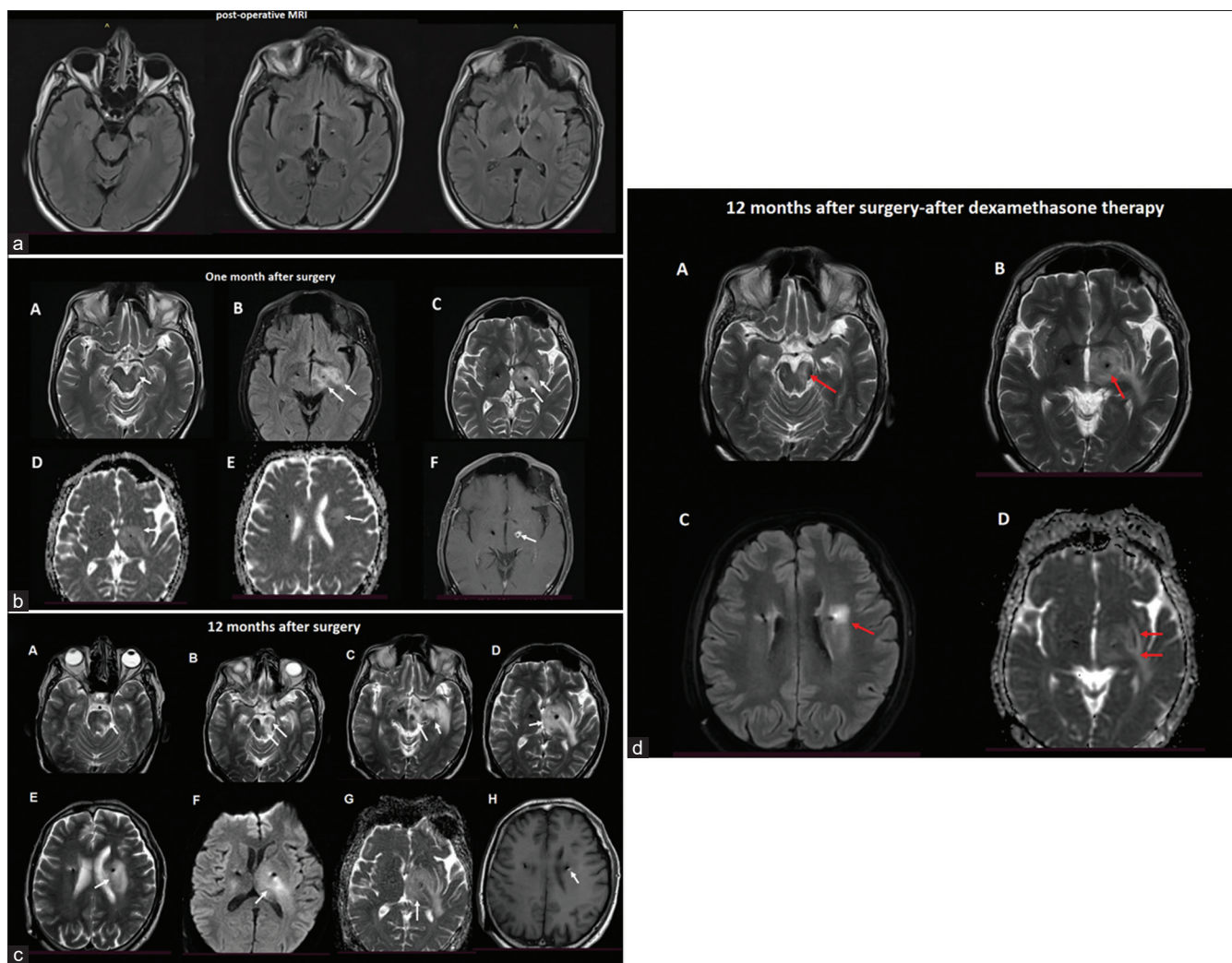


Figure 1: (a) The follow-up postoperative cranial MRI performed 5 days after surgery was within normal limits. (b) The cranial MRI performed 1 month after surgery due to complaints of blurry vision showed left peri-lead vasogenic edema involving the left cerebral peduncle, thalamus, and corona radiata (A–C: T2-weighted images, C–E: Apparent diffusion coefficient maps, F: post-contrast T1-weighted image). (c) The cranial MRI performed 12 months after surgery showed progression in the left peri-lead edema surrounding the left medulla oblongata and basal ganglia (A–E: T2-weighted images, F: Diffusion-weighted image, G: Apparent diffusion coefficient map, H: post-contrast T1-weighted image). (d) The follow-up MRI performed 2 weeks later showed significant resolution of the peri-lead edema (A–C: T2 weighted images, D: Apparent diffusion coefficient map)

In conclusion, we present an illustrative patient developing persistent PLE during the late period of surgery and draw attention to the importance of the recognition of this entity in clinical practice. Prospective neuroimaging studies including long-term follow-up may provide critical contributions regarding the actual frequency and significance of PLE in those patients undergoing DBS therapy.

*: constant voltage stimulation

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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