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Case report

Delayed intracerebral hemorrhage associated with placement of a deep brain stimulating electrode over two years prior

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ABSTRACT

Introduction and importance: Most severe complications of DBS appear in the perioperative period. There are no published case reports of delayed ICH occurring more than three months from electrode implantation. The pathogenesis of delayed ICH remains unclear.

Case presentation: We present a 64-year-old male with essential tremor who sustained a delayed intracerebral hemorrhage (ICH) 2.5 years after implantation of a deep brain stimulating electrode.

Discussion: The patient sustained a thalamic-midbrain ICH that may have been related to the positioning of the electrode. An analysis was performed to determine the cause and risk factors that may have contributed. Based on these findings, it is possible that the proximity of the cannula or electrode may have mildly injured the wall of the superior thalamic vein during implantation, or perhaps being in contact with the vein over a longer-term having an effect, which in either of these scenarios can subsequently lead to ICH formation on the sudden rise of intracranial pressure.

Conclusion: It emphasizes the importance of proper surgical navigation planning, image- guidance, and the use of image verification.

1. Introduction

In the last 30 years, the use of deep brain stimulation has become a standard of care for the treatment of movement disorders [2]. Most commonly, deep brain stimulation (DBS) electrodes are implanted into the ventralis intermedius nucleus (VIM) or zona incerta for the treatment of essential tremor (ET), and either Subthalamic nucleus or Globus Pallidus for Parkinson's disease. Despite the invasiveness of the treatment, it is accepted as a safe and cost-effective treatment for ET that is well- tolerated. Being an elective procedure, it is imperative to maintain a low complication rate. Most serious complications appear in the perioperative period. Procedural-related complications can include infection, as well as subdural or intracerebral hematoma. Post-operative bleeding is commonly noted within the first 48 h after the procedure. The risk of intracerebral hemorrhage (ICH) surrounding the procedure has been reported ranging from 0.2% to 5.6% [9].

Interestingly, there is also an estimated 3.7% incidence for delayed ICH [3]. However, there are no published case reports of delayed ICH occurring more than three months from implantation. We present a case of delayed ICH occurring 2.5 years from DBS electrode implantation in patient with ET. The case report has been reported in line with the SCARE 2020 criteria [1].

2. Case report

A 64-year-old right handed male with a ten year history of worsening right greater than left upper extremity essential tremor. His past medical history was significant for an 8 year history of type II diabetes mellitus, which was controlled with daily Diaprel and Metformin. There was no history of hypertension or anticoagulation therapy. The patient developed severe dysfunction with the tremor and had difficulty with pouring liquids, drinking and eating. Medical treatments included Propranolol but was not tolerated secondary to hypotension with 40 mg daily dosing. Additional treatments without relief included Clonazepam. Work-up included an MRI of the brain, which revealed marked brain atrophy. A

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Abbreviations

ICH	Intracerebral hemorrhage
DBS	Deep brain stimulation
VIM	Ventralis intermedius nucleus
ET	Essential tremor
MER	Microelectrode recording
CT	Computed tomography
MRI	Magnetic resonance imaging

neuropsychological assessment showed a mild cognitive decline (MMSE-26). The decision was made to proceed with DBS. Secondary to the neuropsychological finding of mild cognitive decline, it was decided to perform a unilateral procedure to treat the patient's worse side. The patient underwent placement of a left-sided VIM deep DBS implantation for his ET. The procedure was performed by a functional neurosurgeon with ten years of experience of DBS implantations, using microelectrode recording (MER), and three MER passes were utilized during the procedure. A Medtronic 3387 electrode (Medtronic, Minneapolis, Minn., USA) was implanted. He subsequently underwent general anesthesia for a single-stage procedure with immediate implantation of the implantable pulse generator (Model Activa PC; Medtronic, Minneapolis, Minn., USA). There were no postoperative complications. Immediate postoperative CT of the head demonstrated no hemorrhage or electrode dislocation. After two weeks, stimulation was turned on with complete resolution of right-hand tremor and was satisfied of treatment outcomes. The most dorsal contacts under monopolar stimulation (stimulation parameters 130 Hz, 60 ms, 2,6 V) was utilized. The patient returned for routine follow-up at 3 and 12 months. At his follow-up, there was no evidence of tremor or neurological deficit. Further, follow-up was carried out by family doctor.

He presented to the emergency department with ambulance 2.5 years from his procedure with sudden onset right-sided hemiparesis and unconsciousness. Upon presentation his Glasgow coma scale (GCS) score was six, and he subsequently was intubated. A CT scan of the head demonstrated an acute 1.8 cc left-sided hemorrhage in the thalamus extending into the midbrain [Fig. 1].

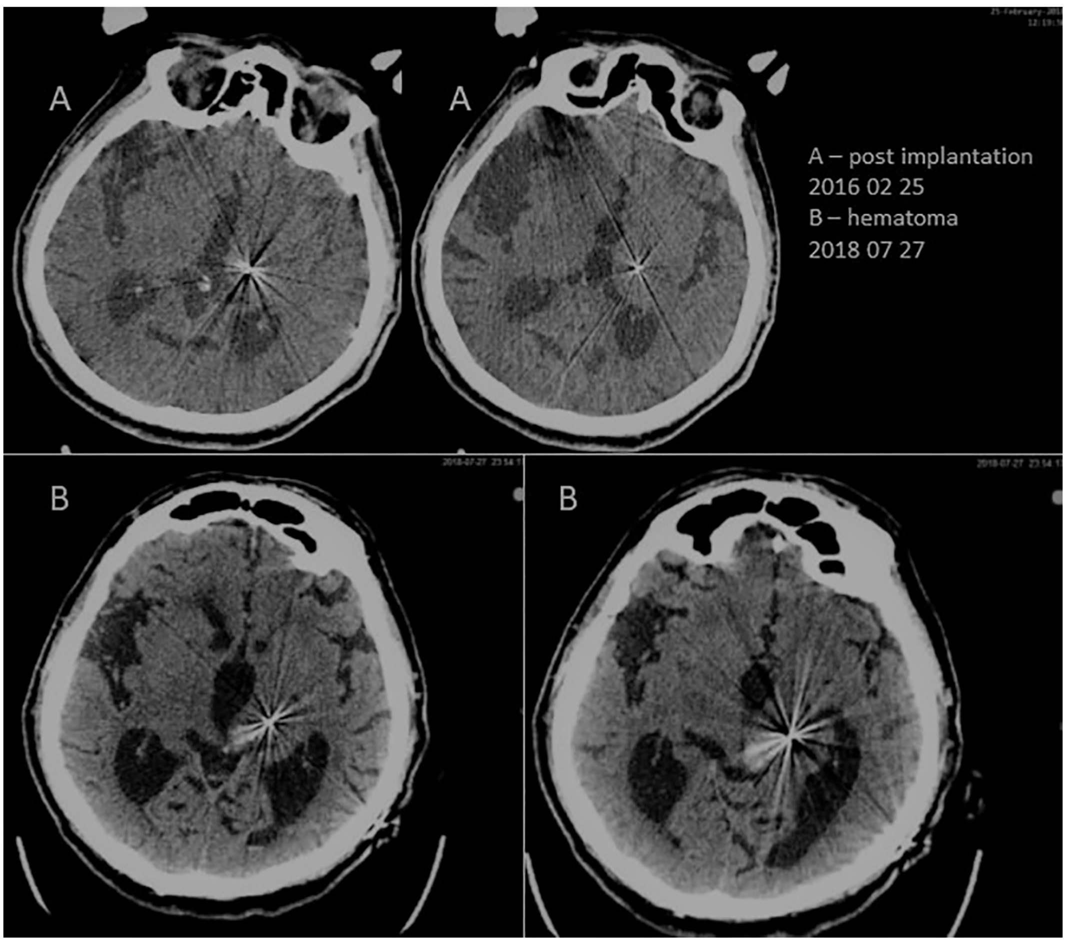

Fig. 1. CT scan intraoperatively after DBS electrode implantation (A) and with ICH after 2.5 years (B).

The hematoma was in contact with the previously implanted Medtronic DBS electrode. The patient was admitted to the neurointensive care unit for observation and treatment. Initial physical examination revealed right-sided hemiplegia with a GCS score of 8. Interestingly, he did not demonstrate diplopia. Blood pressure on admission was 162/82 mmHg. Initial blood tests revealed normal blood counts, urea, glucose, and electrolytes. In addition, clotting factors returned normal. Cardiosonography was negative for possible emboli or occlusion. The patient had improvement in his exam over 48 h and improved to a GCS of 15. His neurologic examination improved to a left leg hemiparesis, with normal upper extremity strength without oculomotor dysfunction or speech disturbance. After two weeks in the hospital, he was transferred to a neurorehabilitation unit and later follow-up was performed regularly by family doctor.

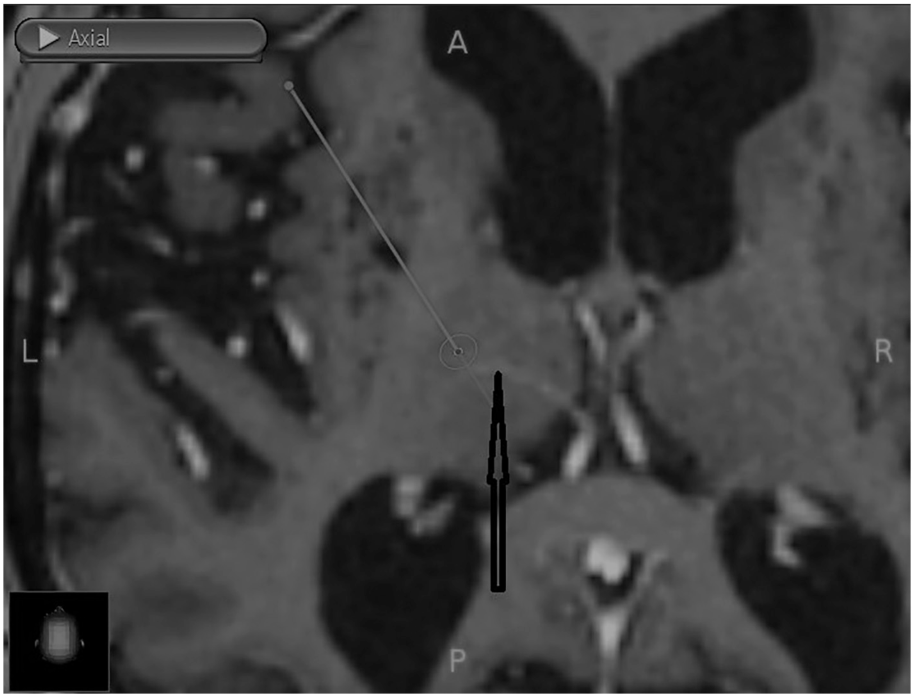

Fig. 2. Superior thalamic vein identified (arrow) during DBS electrode track planning.

3. Discussion

DBS has become a commonly utilized intervention for the treatment of movement disorders. Surgical technique and maintenance of a low complication rate are imperative, being an elective procedure surrounding quality of life. The most severe complication can be an ICH and usually occurs either immediately or within the first 48 h following electrode implantation. Our case report is the first to our knowledge to describe an ICH occurring in a delayed fashion 2.5 years from the implantation. It involved a thalamic-midbrain hemorrhage which lateral border was in contact with the DBS electrode. The risk of intracerebral hemorrhage after DBS electrode implantation ranges from 0.2% to 5.6% [10]. ICH may occur due to direct blood vessel injury. Antiplatelet therapy and hypertension are the main risk factors for such complication [9,10]. There remains debate about the use of MER in increasing the immediate risk of ICH as some have reported an increased risk, while another report demonstrated a decreased risk with multi-channel MER [2,3,9]. The additional data that is obtained by MER in order to accurately place the electrode outweighs the risks of hemorrhage when it is kept significantly low [4,5]. In addition, the literature demonstrates no difference in the average amount of MER passes performed to define the target based on location [6]. Interestingly, there are reports of delayed hemorrhage related to DBS electrode implantation, which had ranged from 2 to 3 months [3,4]. The pathogenesis of delayed ICH remains unclear.

A possible cause of delayed ICH may be spontaneous hemorrhage that occurs irrespective of the implanted DBS electrode. Thalamic hemorrhage counts for up to 8–15% of all ICH and 9.4% for midbrain infarction [7,8].Embolic disease, artery thrombosis, and intrinsic branch penetrator disease are the main pathogenic factors that have been found as the cause. However, in our patient, the cardiosonography was negative for possible emboli or occlusion as the cause of the ICH. In addition, the patient past medical history was negative for hypertension, and their diabetes was very well controlled. This would make a coincidental spontaneous thalamic ICH unlikely.

The thalamus maintains its blood supply by four main branches of the posterior cerebral artery: polar, paramedian thalamic-subthalamic, thalamogeniculate and posterior choroidal arteries. Venous drainage of the thalamus is carried out by the anterior thalamic, thalamostriate, superficial, superior thalamic and basal veins. An analysis of neurological state of the patient and the territory of ICH on imaging leads us to suspect that the ICH was due to the rupture of the superior thalamic vein. Given this finding, we also retrospectively analyzed the preoperative planning and images from SurgiPlan [Fig. 2].

Interestingly, we found the superior thalamic vein in the trajectory of the MER cannulas and final electrode passage. Based on these findings it is possible that the proximity of the canula or electrode may have mildly injured the wall of this vein during implantation, or perhaps being in contact with the vein over a longer term having an effect, which in either of these scenarios can subsequently lead to ICH formation on sudden rise of intracranial pressure.

There has been a review article that concluded venous infarction is the main factor in delayed hemorrhage after DBS surgery [9]. The conclusion was to take care and caution of cortical venous structures during the electrode track planning. Review of the literature and our patient demonstrates that risk factors related to the surgical technique in our case report included the use of MER, and final lead placement. More caution may have been taken in the surgical navigation planning with perhaps a different trajectory and positioning. Based on the findings, it would be difficult to determine if there was increased risk secondary to multiple MER passes, or more so a product of the vicinity of one or all of the passes to the vein. Ultimately, given the timing of a delayed ICH, the culprit may have been the final electrode position. The incidence of hemorrhage in studies adopting an image-guided and image-verified approach was significantly lower than that reported with other operative techniques further demonstrating the importance of navigation and surgical planning [10].

4. Conclusion

This case report illustrates the possibility of a delayed ICH secondary to an implanted DBS electrode over two years from the procedure. DBS candidates should be instructed about delayed ICH risk. It also may strengthen the need for more in depth surgical navigation planning, image- guidance, and the use of image verification.

Ethical approval

The study is exempted from ethical approval.

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CRediT authorship contribution statement

Andrius Radziunas – wrote draft and the final manuscript, also was operating neurosurgeon in this case report

Vytenis Pranas Deltuva – proposed to write a case report and gave critical instructions for manuscript preparation and critically reviewed

Arimantas Tamasauskas – critically reviewed written manuscript

Adomas Bunevicius - critically reviewed written manuscript and made an important change in manuscript

Steven Falowski - critically reviewed written manuscript and made an important change in language and final message of this case report

Guarantor

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Consent

“Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request”.

Declaration of competing interest

The authors report no declarations of interest.

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