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Case Report

Deep brain stimulation of a patient with psychogenic movement disorder

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Abstract

Background: The long-term safety of deep brain stimulation (DBS) is an important issue because new applications are being investigated for a variety of disorders. Studying instances where DBS was inadvertently implanted in patients without a movement disorder may provide information about the safety of the therapy. We report the case of a patient with a psychogenic movement disorder treated with deep brain stimulation (DBS).

Case Description: The patient presented at our clinic after 5 years of chronic DBS of the subthalamic nucleus (STN) for presumed Parkinson's disease. A dopamine transporter (DAT) scan (DaTscan) showed normal DAT distribution in the striatum. A positron emission tomography (PET) scan showed no abnormal metabolic patterns. Further psychiatric and neurological evaluations revealed that the patient was suffering from a psychogenic movement disorder. The patient displayed no sign or symptom from the stimulation, and DBS did not lead to any benefits or side effects for this patient.

Conclusion: We argue that the absence of side effects, the normal DaTscan, and PET scan after 5 years of chronic stimulation illustrate the safety of DBS on neural tissue.

Key Words: Deep brain stimulation, functional imaging, Parkinson's disease, psychogenic movement disorder

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INTRODUCTION

Deep brain stimulation (DBS) of the subthalamic nucleus (STN) leads to symptoms reduction and functional improvements in Parkinson's disease (PD) patients.[9] Although the mechanism of action is not fully understood, DBS may increase the metabolism of STN, as seen on positron emission tomography (PET) scans,[4] and normalize patterns of cerebral activation during movement, as seen on functional MRI (fMRI).[5] The long-term safety of DBS is an important issue given the growing number of patients receiving the treatment and possible new applications in the future. Postmortem studies have confirmed the absence of significant tissue lesion.[1,2] However, the direct effects of DBS on

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neural tissue remain unclear. We present the case of a 60-year-old male suffering from psychogenic movement disorder misdiagnosed as Parkinson's disease and who was implanted with bilateral STN DBS.

CASE REPORT

Our patient initially presented with right upper extremity tremor that eventually generalized to all extremities. He was diagnosed with Parkinson's disease but failed to respond to various medications including levodopa and pramipexole. One year later, he was deemed refractory to medical therapy and underwent the implantation of a DBS system in the STN bilaterally. Five years later, the patient moved to our state and was seen in our clinic.

During our assessment, the patient complained of freezing episodes and dystonic movements wherein he would drool, become aphasic, and experience arm flexion and toe curling. The patient reported that these episodes occurred approximately 3–4 times daily and lasted 5–10 minutes, with no identifiable triggers and no relationship to his medication dosing. DBS did not improve his symptoms, and he had undergone a trial with DBS off for thirty days and noted no significant difference in his symptoms. The patient was taking Sinemet, pramipexole, trihexiphenidyl, Apokyn with no significant benefit. He experienced no benefit from additional medications for presumed dystonic spells including topiramate and rasagiline.

We confirmed the proper placement of DBS leads in the STN bilaterally with magnetic resonance imaging (MRI). The DBS system was programmed for monopolar stimulation using the more ventral contacts with a pulse width of 60 μs, a frequency of 185 Hz, and amplitude of 2.8 V. We obtained a ^{18}F-fluorodeoxyglucose (FDG) PET study of the brain to identify DBS-related alterations in cerebral metabolism that could explain the symptoms [Figure 1]. The left-sided electrode was turned off and the right-sided electrode was kept on. The PET scan showed no asymmetry in cerebral metabolism and no evidence of abnormal FDG uptake. The stimulator was turned off for 1 month without any symptomatic changes.

A dopamine transporter nuclear study (DaTscan) was performed. The study revealed normal distribution of the dopamine transporter within the striatum [Figure 2], which was inconsistent with the diagnosis of any parkinsonian syndromes. Based on these findings and the overall presentation, it was concluded that the patient was suffering from a psychogenic movement disorder. He was referred to a psychiatrist and he is currently being treated for major depressive disorder.

DISCUSSION

Psychogenic movement disorder is a challenging condition, and it can be difficult to differentiate it from a true movement disorder. For instance, a recent study reported on two patients with psychogenic dystonia who underwent DBS. In these cases as well, the patients did not suffer from stimulation-related side effects.[7] Our patient underwent high frequency stimulation of the STN for approximately 5 years without side effects prior to being turned off. Furthermore, his DaTscan showed a normal distribution of dopamine transporters within the striatum. This finding suggests that DBS did not interfere with the distribution of dopamine reuptake receptor within his striatal circuitry. On F-18 FDG PET scanning, we saw neither increased metabolic activity in the lentiform nucleus or thalamus nor decreased activity in the motor and pre-motor cortices, which

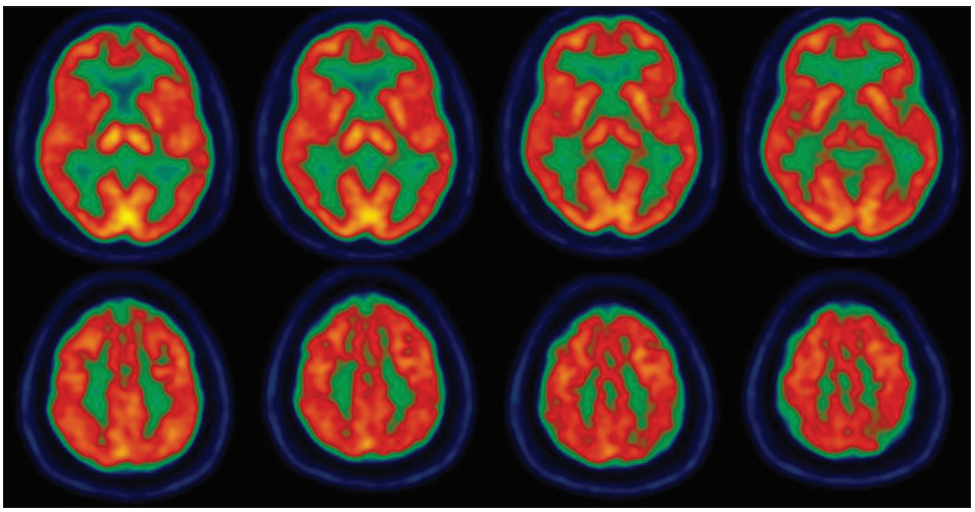

Figure 1: Brain F-18 FDG PET of our patient. The right electrode was ON and the left electrode was OFF during the study. The study shows no difference in metabolic activity between the two hemispheres despite the difference in electrical stimulation

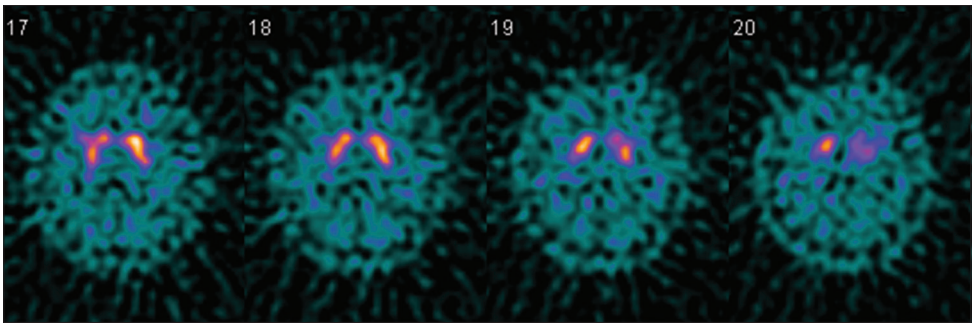

Figure 2: Single photon (SPECT) imaging obtained following the injection of Iodine-123 Ioflupane (DaTscan). The images show a normal distribution of dopamine reuptake transporters at the presynaptic terminals within the striatum bilateral. The findings are inconsistent with Parkinson's disease

have been found even in mild cases of PD.[6] Regional PD related covariance patterns have been reported on FDG PET imaging with DBS in both the ON and OFF state.[3,8] However, we did not see any differences in network activity when comparing the ON hemisphere to the OFF hemisphere in this case. These findings suggest that chronic DBS may not induce metabolic changes in the motor circuits in psychogenic movement disorder. The lack of side effects or stimulation-related effects with long-term DBS in this patient also supports this view.

CONCLUSION

Previous studies have demonstrated the long-term safety of DBS from an anatomical perspective by confirming the absence of significant tissue lesion on postmortem evaluations.[1,2] We now argue that chronic DBS may not affect the metabolism of motor circuits in psychogenic movement disorder. We recognize that this report has several limitations. It presents data from a single patient and may not be representative of a larger cohort. Furthermore, although DBS does not show any alterations in metabolism at this stage, it is still possible that chronic electrical stimulation has brought subtle changes to the neural tissue, such as upregulation or downregulation of receptors that could not be observed on a simple FDG PET or on a DaTscan. The absence of side effects with chronic stimulation is notable and has been reported for psychogenic dystonia as well.

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Conflicts of interest
There are no conflicts of interest.

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