Published Ahead of Print on April 13, 2018 as 10.1212/WNL.0000000000005530

CLINICAL/SCIENTIFIC NOTES

Functional movement disorders arising after successful deep brain stimulation

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Neurology® 2018;0:1-2. doi:10.1212/WNL.0000000000005530

Deep brain stimulation (DBS) is an effective and established treatment for movement disorders, as well as an emerging treatment for psychiatric disorders. Outcome is influenced by many factors, including patient selection, electrode placement, stimulation parameters, hardware issues, oral medication adjustments, disease progression, and comorbidities. We present another potential complication following DBS surgery that is clinically important and probably underreported—the onset of a new functional movement disorder. In each of the 4 cases described (all of whom were treated at Toronto Western Hospital over a 15-year period), the correct diagnosis led to diagnostic counseling including demonstration of the presence and significance of positive functional signs to the patient. Patient 4 also had a short course of physiotherapy. Resolution of functional symptoms occurred within 3 months in all cases.

Case series

Patient 1

A 51-year-old woman with Parkinson disease (PD) underwent staged bilateral pallidotomy 10 years after disease onset due to severe levodopa-induced dyskinesias. Eight years later, bilateral DBS of the subthalamic nucleus (STN) was performed due to the re-emergence of disabling motor fluctuations, which were successfully treated. Comorbid anxiety and social phobia predated the DBS surgery. The early postoperative management was complicated by the sudden onset of functional kicking out of the right leg triggered by the act of sitting down (video 1, links.lww.com/WNL/A453).

Patient 2

A 67-year-old woman with PD underwent bilateral STN DBS 9 years after disease onset due to severe motor fluctuations and off-state dystonia. There was a 50% improvement in Unified Parkinson's Disease Rating Scale part III score. Ongoing right upper limb rest tremor and right foot dystonia improved with increasing simulation voltage and botulinum toxin, respectively. On multiple occasions, she mentioned that her family had left her alone after surgery. She subsequently developed a new functional right leg tremor that was variable, distractible, and more evident when she was under the false impression that the stimulation had been switched off (video 2, links.lww.com/WNL/A454).

Patient 3

A 21-year-old woman with generalized dystonia due to neonatal hypoxic brain injury underwent bilateral DBS of the globus pallidus pars interna (GPi). Comorbid anxiety and emotional lability predated the DBS surgery. Burke-Fahn-Marsden Dystonia Rating Scale severity/disability scores declined from 24.5/8 preoperatively to 11.5/5 at 12 months. One year later, she became frustrated that she "would never be normal." She acutely developed functional flexion-extension movements of the right wrist and fingers that were distractible and suggestible (video 3, links.lww.com/WNL/A455).

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Patient 4

A 37-year-old man underwent bilateral DBS of the subcallosal cingulate gyrus (Cg25) for treatment-resistant depression as part of an experimental trial.[1] Subsequent to this and other pilot studies, a larger randomized trial reported no significant antidepressant efficacy of Cg25 stimulation during the double-blind, sham-controlled phase.[2] His clinical course was complicated by the need for system reimplantation at 1 and 10 years due to hardware infection; however, on each occasion, his depressive symptoms improved significantly after surgery. He had comorbid chronic whole body pain. After the second implant, he began to complain of functional leg weakness, balance impairment, and tremor (that was variable in frequency and distractible) in the absence of depressive symptoms (video 4, links.lww.com/WNL/A456).

Discussion

Functional diagnoses are a contraindication for DBS, but some patients have undergone an operation as a result of misdiagnosis and this is a recognized cause of surgical failure.[3] In this article, we describe 4 patients in whom functional movement disorders arose only after successful DBS for various conditions (table e-1, links.lww.com/WNL/A452), a situation resembling the appearance of nonepileptic attacks in patients following epilepsy surgery.[4] Patients frequently struggle to psychologically adapt to the rapid improvement in their function and health after DBS surgery.[5,6] This difficulty in renouncing the sick role could potentially contribute to the development of functional symptoms, although the delayed symptom onset following DBS implantation in some of our patients may point to other factors being involved (such as unmet expectations, comorbid psychiatric problems, medication reduction, altered body image, and direct stimulation effects on limbic or other pathways). In the rare cases of suicide following DBS—which some but not all studies have suggested may be more common than in the general population—some of these factors may also be contributory.

Functional movement disorders arising after DBS have previously been reported in single patients with essential tremor[7] and Tourette syndrome.[8] Functional movement disorders should be considered as a potential cause of poor DBS outcome and are likely to be underrecognized, although further studies involving other high volume DBS centers should be done to substantiate this. In keeping with the existing literature, our case series supports the view that early recognition of functional motor disorders increases the likelihood of a good recovery.[9] It also reinforces the idea that a functional disorder can emerge at any time and that surgical procedures have the potential to blind the clinician to their occurrence.

Author contributions

Dr. Breen wrote the manuscript. Dr. Rohani, Dr. Moro, Dr. Mayberg, Dr. Zurowski, and Dr. Lozano managed the patients and revised the manuscript. Dr. Fasano conceived the idea, managed the patients, and revised the manuscript.

Study funding

D.P.B. is funded by an Edmond J. Safra Fellowship in Movement Disorders via the Michael J. Fox Foundation.

Disclosure

D. Breen and M. Rohani report no disclosures relevant to the manuscript. E. Moro has received honoraria from Medtronic and grant support from the CHU of Grenoble, the Grenoble Alpes University, and Merz. H. Mayberg receives consultancy fees and intellectual property licensing fees from St Jude Medical. M. Zurowski reports no disclosures relevant to the manuscript. A. Lozano receives consultancy fees from Medtronic, St Jude Medical, and Boston Scientific and holds the RR Tasker Chair in Functional Neurosurgery and Tier 1 Canada Research Chair in Neuroscience. A. Fasano receives consultancy fees from AbbVie, Medtronic, Boston Scientific, Sunovion, Chiesi farmaceutici, UCB, and Ipsen; receives honoraria from AbbVie, Medtronic, Boston Scientific, Sunovion, Chiesi farmaceutici, UCB, and Ipsen; receives grant support from University of Toronto, Weston Foundation, AbbVie, Medtronic, and Boston Scientific; and sits on advisory boards for AbbVie, Boston Scientific, and Ipsen. Go to Neurology.org/N for full disclosures.

Received November 4, 2017. Accepted in final form February 12, 2018.

References

1. Mayberg HS, Lozano AM, Voon V, et al. Deep brain stimulation for treatment-resistant depression. Neuron 2005;45:651–660.
2. Holtzheimer PE, Husain MM, Lisanby SH, et al. Subcallosal cingulate deep brain stimulation for treatment resistant depression: a multisite, randomised, sham-controlled trial. Lancet Psychiatry 2017;4:839–849.
3. Pauls KAM, Krauss JK, Kämpfer CE, et al. Causes of failure of pallidal deep brain stimulation in cases with pre-operative diagnosis of isolated diagnosis. Parkinsonism Relat Disord 2017;43:38–48.
4. Krahn LE, Rummans TA, Sharbrough FW, et al. Pseudoseizures after epilepsy surgery. Psychosomatics 1995;36:487–493.
5. Schüpbach M, Gargiulo M, Welter ML, et al. Neurosurgery in Parkinson disease: a distressed mind in a repaired body? Neurology 2006;66:1811–1816.
6. Houeto JL, Mallet L, Mesnage V, et al. Subthalamic stimulation in Parkinson disease: behavior and social adaptation. Arch Neurol 2006;63:1090–1095.
7. McKeon A, Ahlskog JE, Matsumoto JY. Psychogenic tremor occurring after deep brain stimulation surgery for essential tremor. Neurology 2008;70:1498–1499.
8. Duits A, Ackermans L, Cath D, et al. Unfavourable outcome of deep brain stimulation in a Tourette patient with severe comorbidity. Eur Child Adolesc Psychiatry 2012;21:529–531.
9. Gelauff J, Stone J, Edwards M, Carson A. The prognosis of functional (psychogenic) motor symptoms: a systematic review. J Neurol Neurosurg Psychiatry 2014;85:220–226.

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