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Case study

Successful deep brain stimulation for central post-stroke pain and dystonia in a single operation

Marshall T. Holland [a], Mario Zanaty [a], Luyuan Li [a], Teri Thomsen [b], James H. Beeghly [c], Jeremy D.W. Greenlee [a,*], Chandan G. Reddy [a]

[a] Department of Neurosurgery, University of Iowa Carver College of Medicine, Iowa City, IA, United States
[b] Department of Neurology, University of Iowa Carver College of Medicine, Iowa City, IA, United States
[c] Department of Psychiatry, University of Iowa Carver College of Medicine, Iowa City, IA, United States

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ABSTRACT

Background: Central post-stroke pain is known to be refractory to medications and difficult to manage. We present a case of central post-stroke pain associated with dystonia. Both conditions were successfully treated with a single deep brain stimulation (DBS) operation.

Case Description: A 60-year-old female suffered a right posterior cerebral artery stroke following emergent clipping of a ruptured posterior cerebral artery aneurysm resulting in central post-stroke pain. This manifested as delayed left face and hemibody allodynia and hyperesthesia. The patient also developed marked left-sided dystonia. These progressive symptoms were disabling and refractory to conservative management. The patient underwent a single-stage DBS surgery with stereotactic targeting and implantation of two leads. One lead was placed in the right-sided ventral capsule/ventral striatum for treatment of pain and a second lead in the right-sided globus pallidus interna for treatment of dystonia.

The surgical implantation proceeded without complication. The patient's dystonia markedly improved following surgery. While her pain improved, she required multiple, meticulous programing sessions to achieve significant pain relief and decrease in pain medication use. Overall, the patient was satisfied with the results of her intervention. She did, however, have occasional intermittent spells of severe pain on top of her residual discomfort throughout her treatment course. Unfortunately, she died from small cell lung carcinoma a year after her DBS surgery.

Conclusions: Deep brain stimulation targeting multiple brain networks in one operation is feasible and safe. Deep brain stimulation may be considered in some refractory cases of central post-stroke pain; however, it requires meticulous programming.

1. Introduction

Pain of central origin is notorious for being refractory to medical treatment, as documented in several studies [1–3]. Central post-stroke pain (CPSP) is characterized by allodynia or hyperesthesia that is chronic with associated continuous numbness typically involving one side of the face and hemibody. The pain is unrelenting, refractory, and extraordinarily frustrating for patients and treating physicians. It occurs in approximately 8% of all stroke patients but may be as high as 18% in those with sensory deficits [4]. While a prior case abstract reported improvement of tremor following DBS of the periventricular grey matter (PVG) and ventroposteriolateral thalamic nucleus (VPL), to our knowledge we present here the first report of a post-stroke patient with CPSP and co-morbid dystonia treated with a single deep brain stimulation procedure with one lead implanted in the globus pallidus interna (GPi) for dystonia and one lead placed in the ventral capsule/ventral striatum (VC/VS) for pain [5].

2. Case description

A 60-year-old female first presented to the neurosurgical service with acute subarachnoid hemorrhage due to a ruptured right posterior cerebral artery aneurysm. She underwent emergent

Abbreviations: AC-PC, anterior commissure-posterior commissure; DBS, deep brain stimulation; GPi, globus pallidus interna; Hz, Hertz; IPG, impulse generator; MCP, mid-commissural point; PCA, posterior cerebral artery; PVG, periventricular gray region; CPSP, central post-stroke pain; V, volts; VC/VS, ventral anterior internal capsule/ventral striatum; VS, ventral striatum; μs, microseconds.

* Corresponding author at: Department of Neurosurgery University of Iowa Hospitals and Clinics, 200 Hawkins Drive, Iowa City, IA 52242, United States.
E-mail address: jeremy-greenlee@uiowa.edu (J.D.W. Greenlee).

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microsurgical clipping of the aneurysm. This procedure was complicated by a right posterior cerebral artery stroke that involved her thalamus. (Fig. 1) Postoperatively, she had residual deficits including a dense left homonymous hemianopsia, hemiparesis, and numbness of the face and hemibody. Seven years after the initial stroke, she began to develop left-sided pain, allodynia, and hyperesthesia involving her face and hemibody that were previously numb. With the development of the pain came associated dystonia. Her pain was progressive and refractory to medical management. She required multiple classes of pain medication, including amitriptyline (which helped with the pain but led to hypertension), baclofen, gabapentin, carbamazepine, oxcarbazepine, duloxetine, pregabalin, and propoxyphene. She was significantly affected by her pain. This hampered her post-stroke recovery and rehabilitation, resulting in a depressed mood with social isolation. She experienced intermittent insomnia, decreased ability to concentrate, and mild anhedonia. Despite these experiences she did not meet the diagnostic criteria for depression, anxiety, or post-traumatic stress disorder that would require formal psychiatric treatment. Ultimately, she was diagnosed with psychological factors affecting a physical illness. After her case was discussed at a multidisciplinary conference including neurosurgery, psychiatry, and pain colleagues, IRB approval was obtained for placement of a deep brain stimulation (DBS) lead in the VC/VS for her pain. The patient decided to proceed with DBS to address both her dystonia and pain syndrome.

2.1. Surgical technique

The patient underwent right-sided DBS lead (model 3391, Medtronic, Minneapolis, MN) implantation in the VC/VS for pain and right-sided GPi lead (model 3387, Medtronic, Minneapolis, MN) implantation for dystonia. The GPi target was chosen based on the extensive literature showing improvements of focal and generalized dystonia with GPi DBS [6]. VC/VS was selected based on reports of improved pain [7] and depression [8]. Our center has considerable experience with GPi targeting for the treatments of Parkinson' s disease and dystonia and VC/VS targeting for refractory obsessive-compulsive disorder under IRB approvals. Prior to surgery, a high-resolution volumetric brain MRI was obtained (1-mm slices). The morning of surgery, the patient was brought into the operating room, placed under general anesthesia, placed in a stereotactic ring (CRW, Integra, Plainsboro, NJ) and a volumetric CT scan was obtained. The two scans were merged for targeting and trajectory planning on a workstation (Stealth, Medtronic, Minneapolis, MN). Direct targeting was used to obtain coordinates for the VC/VS on the right side and were deduced as 9 mm lateral, 11 mm anterior, and 4 mm deep to mid-commissural point (MCP) of the anterior commissure-posterior commissure (AC-PC) line. A trajectory was chosen so that the electrode traveled down the anterior limb of the internal capsule. Indirect targeting was used to obtain starting coordinates for the right GPi nucleus and were 21 mm lateral, 2 mm anterior, and 4 mm deep to MCP. Planned incisional markings were placed at 3.5 cm and 5.3 cm lateral to the midline and along the coronal suture for the GPi and VC/VS burr holes respectively. Both trajectories were planned to avoid prominent sulci, ventricles, and the head of the caudate. Intraoperative physiology was used to determine ultimate lead positioning for each target. For VC/VS, macrostimulation was performed. At target depth, a transient increase of heart rate of 10 beats per minute was noted with stimulation of the deepest contact (i.e. contact 0) and no adverse effects were evident with stimulation up to 5 V. For GPi target refinement, microelectrode recordings were obtained under propofol and remifentanil anesthesia and defined the superior and inferior borders of GPi. No photic response was noted. The DBS lead was placed with contact 0 at the inferior border of GPi. Macrostimulation was performed and revealed a transient release of dystonia in the left hand without evident side effects up to 5 V. Following implantation of both leads, the stereotactic frame was removed, the patient repositioned, and an impulse generator (Activa PC, Medtronic, Minneapolis, MN) was placed in the right chest and connected to the electrodes with extension wires. The operation proceeded without complication and the patient was able to be discharged home on postoperative day 2. Postoperative imaging is shown in Fig. 2.

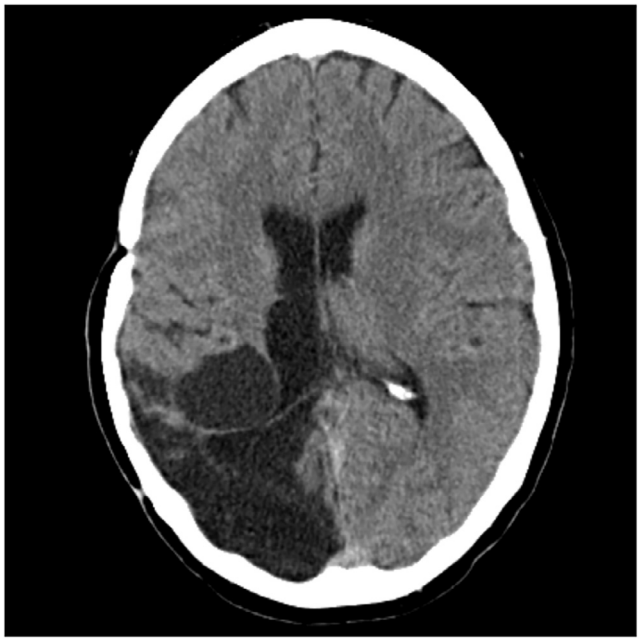

Fig. 1. Right Posterior Cerebral Artery Stroke Axial head CT without contrast obtained 6 years after neurosurgical clipping of right posterior cerebral artery (PCA) showing diffuse hypodensity and encephalomalacia in the right PCA vascular territory.

2.2. Post-surgical course

Immediately after surgery there was a micro-lesional effect noted with some improvement of the patient's dystonia prior to stimulator activation. The following day, the patient underwent initial programming by a fellowship-trained movement disorder neurologist highly experienced in DBS. The dystonia responded well to GPi DBS with improvements in her ability to walk with the use of a left leg straightener and decreased left sided toe curling. The patient reported decreased tightness on her left side. On the other hand, her pain was significantly more difficult to control with VC/VS DBS. Eventually, after multiple meticulous programming sessions the patient did find relief and was able to reduce her pain medication requirements. Her pain visual analogue scores improved from 8 out of 10 pre-operatively to 5 out of 10 six months post-operatively. Her final stimulation settings are summarized in Tables 1 and 2. Despite the overall improvement in her pain, she continued to have intermittent bouts of severe pain and some residual discomfort. The patient also reported significant improvement in her mood and she was also able to successfully reintegrate into society. During a postoperative visit with her psychiatrist, the patient denied having any depressive symptoms or alterations in mood. Objectively, her Inventory of Depressive Symptomatology score decreased from 13 preoperatively to 2 two months postoperatively. Her affect pain score from the McGill Pain Questionnaire-2 decreased from 34 preoperatively to 10 over the course of one year. She was overall satisfied with the results,

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