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CASE REPORT

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Effective treatment of refractory complex facial pain with motor cortex stimulation by spinal paddle electrodes using multimodal imaging

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Abstract

Background: Complex facial pain is a debilitating condition with varying etiologies that overall responds poorly to both medical and traditional surgical management. Cortical stimulation is a unique therapeutic intervention which can be effective for some types of complex facial pain syndromes (CFPS). However, the novel use of preoperative functional magnetic resonance imaging (fMRI) and diffusion tensor imaging (DTI) coupled with intraoperative stimulation mapping and phase reversal to improve the accuracy for placement of spinal paddle electrodes in motor cortex stimulation, to our knowledge, has not been reported in the literature.

Case presentation: Here, we present a unique case of a 56-year-old male who developed left-sided complex facial pain syndrome after a stroke refractory to medical management and peripheral nerve stimulation. He previously underwent microvascular decompression (MVD) with limited control of his left-sided facial pain. In order to treat this, the patient underwent motor cortex stimulation. The motor strip of the face and tongue was identified preoperatively with functional magnetic resonance imaging (fMRI) and diffusion tensor imaging (DTI). Intraoperatively, phase reversal was used to identify corticospinal tracts and stimulus mapping confirmed the location before the epidural placement of two spinal paddle electrodes. Postoperatively, the patient reported significant reduction in pain levels, burning dysesthesias, and intensity and frequency of symptoms. This trend continued, and the patient experienced equivalent levels of relief at 6 months.

Conclusions: This is a rare case report of successful motor cortex stimulation with the novel preoperative use of fMRI and DTI, coupled with intraoperative functional mapping, to successfully guide the placement of spinal paddle electrodes for the treatment of CFPS.

Keywords: Neurosurgery, Neuromodulation, Pain, Trigeminal nerve pain, Functional neurosurgery, Neuropathic pain

Background

Motor cortex stimulation (MCS) is a neuromodulating intracranial technique used for treating intractable complex facial pain. This intervention is usually reserved for refractory complex facial pain syndrome (CFPS) patients, where intracranial electrodes are implanted for motor cortex stimulation, identified through functional imaging, phase reversal, and stimulation imaging [1]. Previous observations and animal models have demonstrated that cortical stimulation of the motor cortex inhibits pain through non-nociceptive somatosensory neurons that inhibit nociceptive neurons corresponding to the area of

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facial muscle contraction. The exact mechanism of action is poorly understood. It is hypothesized activation of the primary motor cortex acts as both an ascending and descending modulator. In the descending pathway, MCS activates the periaqueductal gray (PAG), striatum, and cingulate gyrus, thereby regulating opioid and gamma-aminobutyric acid (GABA) levels through serotonin and dopamine release to decrease thalamic activity. The ascending pathway suppresses ectopic discharges from the dorsal horn and regulates tactile thresholds by suppressing neuronal activation in the thalamus from the spinothalamic tract [2–5]. Even with this understanding, the reported clinical outcomes of MCS have been variable [4]. For these reasons, MCS has not been approved by the United States Food and Drug Administration and is considered a last resort option for refractory facial pain syndromes. Thus, limited device options are available, and there is currently no accepted standardized technique.

Here, we describe a 56-year-old male presenting with complex facial pain syndrome refractory to medical management, microvascular decompression, and peripheral nerve stimulation who reported significant improvement after motor cortex stimulation. Novel use of functional magnetic resonance imaging (fMRI), diffusor tensor imaging (DTI), phase reversal, and stimulation mapping was utilized to localize the motor cortex corresponding to the pain region before the epidural placement of spinal paddle electrodes, enhancing successful lead placement.

Case presentation

A 56-year-old male with history of congenital external auditory canal atresia status post multiple reconstructive surgeries presented with severe, burning facial pain in his left cheek and jaw. The pain radiated to his left ear and was associated with intermittent swelling. On examination, the patient demonstrated decreased sensation on the left face in divisions V1–V3, allodynia, and a slight left facial droop. He had previously been diagnosed with trigeminal neuralgia, failing extensive medical management due to poor relief and lack of medication tolerance, which included carbamazepine, gabapentin, antidepressants, and baclofen. He also tried multiple migraine medications and botox to rule out migraines. Six months after failing a trigeminal nerve block, he underwent microvascular decompression at another facility with mild improvement of his facial pain. Postoperatively, he suffered a stroke, resulting in both mild weakness and complex regional pain syndrome involving the left side of his body, including worsening of his facial pain. His pain was unresponsive to non-steroidal anti-inflammatory drugs, steroid injections, multiple antiepileptics, and intravenous opioids and on average rated his pain as 10/10 in severity via the visual analog scale (VAS). He was diagnosed with complex facial pain syndrome. After failure of both peripheral nerve stimulation and transcranial magnetic stimulation, motor cortical stimulation was offered as an off-label therapy for refractory CFPS.

Preoperatively, fMRI and DTI (General Electric Company, Discovery™ MR750w GEM 3.0T, manufactured by General Electric Company, USA; NordicNeuroLab Advanced fMRI Solution, manufactured by NordicNeuroLab Inc., Norway) were used to identify regions within the motor cortex corresponding to tongue and hand regions. The patient was positioned in the supine position, and neuronavigation (Synaptive BrightMatter Guide (V 1.5.4) and Modus Plan (V 2.0.1), manufactured by Synaptive Medical, Canada) was utilized to delineate the hand, face, and tongue regions of the motor cortex using preoperative imaging, shown in Fig. 1. A curvilinear incision was made on the left side of the scalp which incorporated prior incisions. A bone flap was turned and the dura exposed. Phase reversal using a 6-electrode array was used to identify corticospinal tracts, and a stimulus

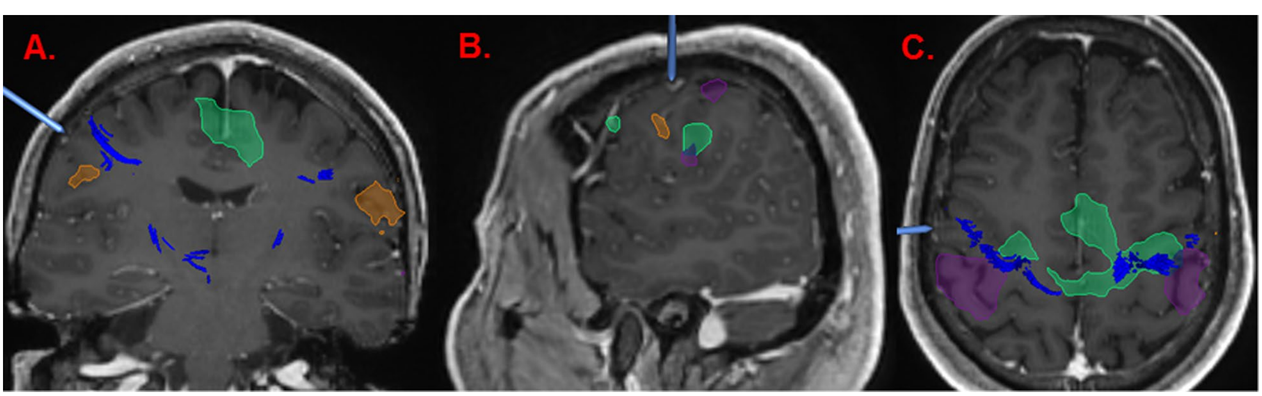

Fig. 1 Preoperative functional MRI. Green = foot, purple = hand, orange = tongue: **A** coronal, **B** sagittal, **C** axial

probe confirmed the location of the motor areas of the hands and face (Cadwell Cascade Pro IONM, manufactured by Cadwell Industries Inc., USA). Once identified, two Abbot paddle spinal cord stimulation electrodes (Abbott Lamitrode 44, Paddle Lead Kit 3244, manufactured by Abbott Laboratories, USA) were placed on the dura over regions corresponding to the motor cortex on fMRI, DTI and cortical mapping (Fig. 2). The skull flap was replaced, and the incision was closed. Postoperative head computed tomography (CT) (General Electric Company, Discovery CT750 HD, manufactured by General Electric Company, USA) was overlaid with the preoperative fMRI, which demonstrated excellent electrode placement (Fig. 3). His postoperative exam was similar to his preoperative baseline. He was initially programmed at a continuous frequency of 50 Hz, pulse width of 250 μs

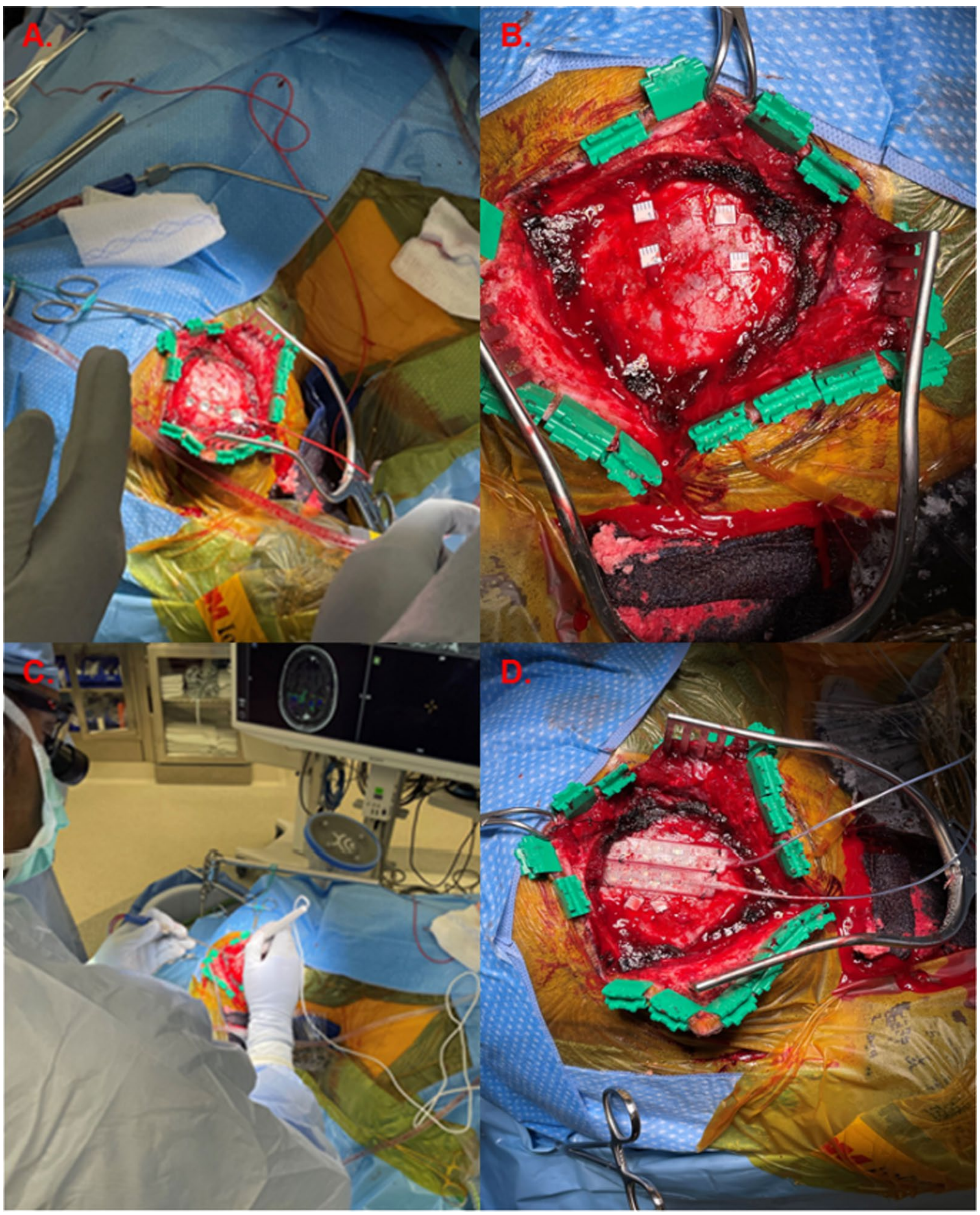

Fig. 2 Intraoperative imaging: **A** epidural phase reversal; **B** epidural markers of facial and hand motor cortex as identified by intraoperative DTI/fMRI; **C** cortical stimulation with EMG/muscle activity proxy; **D** final positioning of epidural paddles

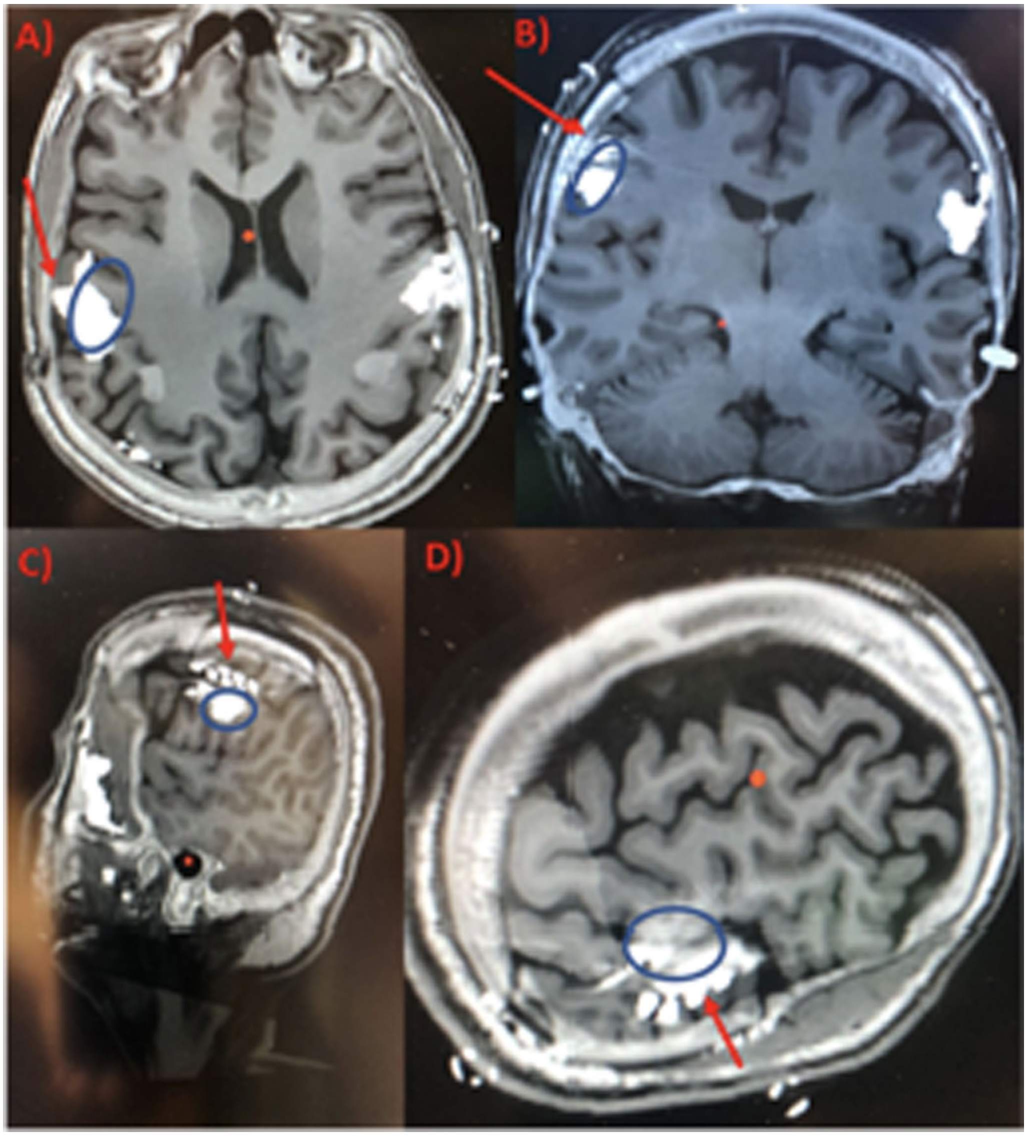

Fig. 3 Postoperative CT overlay with preoperative fMRI. Red arrow is grid, blue circle is tongue functional motor cortex: **A** axial, **B** coronal, **C** sagittal, **D** oblique

at 0.5–11 mA. Upon discharge, the patient reported significant reduction in dysesthesias and allodynia. Two months postoperatively, he complained of worsening pain. Stimulator adjustments were made for alternate programming, which allowed him to increase his continuous dose and pulse width to 60 Hz and 400 µs, respectively, or switch to an intermittent dose of 40 Hz at an intraburst rate of 500 Hz with a pulse width of 1000 µs and maximum current of 1 mA. The patient had some fluctuation of his symptoms with sporadic severe pain attacks which brought him to the emergency department, however, the number of these visits decreased by over 50% compared to his presentation preoperatively. Overall, at 6 months postoperatively, the patient rated his pain on average as 4/10 via VAS, a 60% decrease in severity from his initial presentation.

Discussion

Tsubokawa and colleagues first described MCS in the 1990s, and despite our improved understanding of this technique over the last three decades, it is still considered experimental and is only used as a last resort intervention for refractory CFPS [6]. Aside from its variable outcomes in both dated and current studies, there is an overall lack of evidence-based trials to recommend inclusion and exclusion criteria for MCS. This report demonstrates that epidurally placed spinal paddle electrodes for MCS using functional preoperative and intraoperative stimulation mapping is safe and effective for the treatment of refractory complex facial pain syndrome.

The most crucial aspect of this intervention is the preoperative planning and corresponding intraoperative neuronavigation in order to optimize targeting of MCS. The codependent use of fMRI and intraoperative epidural cortical mapping has been shown to significantly improve the quality of functional targeting of MCS [7]. Furthermore, DTI is a widely accepted, non-invasive tractography tool that allowed us to better visualize the white matter cytoarchitecture before stimulation. As DTI uses fractional anisotropy, which is highly sensitive to changes in cerebral microstructure, we were able to create a 3-dimensional computer model of the patient's fiber tracts that aided our intraoperative navigation [8]. Since appropriate MCS targeting is essential for maximal pain relief, having high-specificity imaging modalities preoperatively was proven to be very useful in our patient. Intraoperative technique is also a critical component during MCS, with phase reversal being performed in order to accurately identify the central sulcus and subsequently place the electrodes in the motor area corresponding to the patient's pain [9]. In addition, stimulation mapping is a cost-effective, easy to use, and readily available modality that actively disrupts a particular function in order for the neurosurgeon to identify the relevant eloquent cortex [10]. All in all, the utilization of multimodality imaging and mapping to confirm the motor areas of interest played a worthwhile and integral role in our achievement of a good patient outcome, allowing us to achieve a 60% reduction in pain severity.

In the original MCS study, Tsubokawa and colleagues used plate electrodes, and many of the later studies of MCS have followed suit [4, 6]. We have found that use of spinal paddle electrodes in MCS increased the ease of manipulation to accurately place and anchor the electrodes to our location of interest. Kolodziej and colleagues also found that the use of spinal paddle electrodes allows for the option of minimally invasive placement through the use of a burr hole, which they found decreased the risk of epidural hematoma, compared to a small craniotomy [11]. We opted to perform a small craniotomy which allowed us to gain a more detailed intraoperative functional map to maximize the accuracy of our electrode placement and increase our area of stimulation using two paddle electrodes.

Henssen and colleagues performed a meta-analysis to assess the efficacy of MCS in chronic neuropathic orofacial pain syndromes. Among 140 patients, they reported that the median pain relief was 64.8% and over 40% pain relief was reported in 68.9% of patients. Although MCS lacks the long-term follow-up of other pain interventions such as microvascular decompression, the gold standard surgery for trigeminal neuralgia, the results are promising, rivaling treatment of CFPS using peripheral nerve stimulation (PNS) [4]. Infection rates are similar in PNS and MCS, however, MCS still presents a larger risk compared to other pain interventions due the involvement of implants within the central nervous system (CNS). Implants also invariably increase the risk of seizures, making this procedure a last resort option [4]. Interestingly, MCS is most effective in treating refractory patients who lack trigeminal nerve lesions such as trigeminal nerve pain, supranuclear lesions including post-stroke pain, and patients without motor deficits [4]. These findings are consistent with this case as our patient presented with CFPS secondary to both trigeminal neuralgia and post-stroke pain.

Conclusions

We report excellent improvement in refractory CFPS symptoms with MCS. Here, the placement of epidural spinal paddle electrodes using preoperative DTI and fMRI coupled with intraoperative functional mapping were safe and efficacious for the patient. Overall, with optimal preoperative planning and intraoperative neuronavigation, this is a promising treatment option for refractory CFPS. We recommend future prospective trials be performed to support its standardized use.

Abbreviations

CFPS: Complex facial pain syndrome; fMRI: Functional magnetic resonance imaging; DTI: Diffusion tensor imaging; MVD: Microvascular decompression; MCS: Motor cortex stimulation; PAG: Periaqueductal gray; GABA: Gamma-aminobutyric acid; VAS: Visual analog scale; CT: Computed tomography; PNS: Peripheral nerve stimulation; CNS: Central nervous system.

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None.

Authors' contributions

Conception and design: MS, JL, AG, LM, DD. Data acquisition: AG, LM, DD. Drafting the manuscript: AG, OG, LM, JL, MS. Critically revising and reviewing the submitted version of the manuscript: all authors (AG, OG, LM, DD, JVD, OA, JL, MS). All authors read and approved the final manuscript.

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Availability of data and materials

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Declarations

Ethics approval and consent to participate

Institutional review board approval was deemed unnecessary. Consent was obtained from patients or their family members before procedures were performed.

Consent for publication

Consent was obtained from the patient or family members before procedures were performed or published.

Competing interests

The authors declare that they have no competing interests.

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