PAIN MEDICINE CASE REPORTS

ISSN 2768-5152

Volume 5, Number 4, pp. 193-196

Successful Treatment of Occipital Neuralgia in a Pediatric Patient with the Use of Peripheral Nerve Stimulator: A Case Report

Allison N. Heizelman, MD[1], Timothy W. Casias, MD[2], Peggy Y. Kim, MD[3], and Benjamin J. Walker, MD[2]

Background: Neuromodulation is a commonly used technique in adult pain management, with current off-label use of peripheral nerve stimulators for the treatment of occipital neuralgia. This targeted therapy can help avoid systemic medications and treat refractory symptoms.

Case Report: We present a 17-year-old boy with significant lesser occipital neuralgia and hydrocephalus status post ventriculoperitoneal shunt placement. He had occipital neuralgia treatment failures with both medication trials and surgical decompression in conjunction with physical and psychological therapies, eventually requiring weekly lesser occipital nerve blocks for pain relief. Our patient experienced a substantial reduction in pain and increase in functional recovery after the placement of a permanent peripheral nerve stimulator without disruption of his ventriculoperitoneal shunt.

Conclusion: Peripheral nerve stimulator implantation can be safe and efficacious for the treatment of refractory occipital neuralgia in a pediatric patient with an ipsilateral ventriculoperitoneal shunt.

Key words: Peripheral nerve stimulation, occipital neuralgia, pediatric, chronic pain, neuromodulation

BACKGROUND

The International Neuromodulation Society defines neuromodulation as "the alteration of nerve activity through targeted delivery of a stimulus, such as electrical stimulation or chemical agents, to specific neurologic sites in the body" (1). Neuromodulation has seen a significant increase in utilization over the last 20 years in adults for various headache indications, including chronic migraine and occipital neuralgia (2).

Occipital neuralgia (ON) involves paroxysmal shooting or stabbing headaches in the distribution of the occipital nerve on the posterior scalp (3). Symptoms can be debilitating, highlighting the need for efficacious treatments. A variety of interventions are currently being used, although there is no clear consensus on recommended management. Initial treatment will often include non-pharmacological interventions (massage, craniocervical exercises, and physiotherapy to improve posture) and pharmacologic management, including nonsteroidal anti-inflammatory drugs, tricyclic antidepressants, muscle relaxants, and anticonvulsants (4). Interventional management includes local anesthetic injection with or without steroids, botulinum toxin infiltrations, pulsed or conventional radiofrequency ablation, and even more invasive surgical management including neurolysis, nerve decompression, or other destructive surgeries (4). Psychological support, biofeedback therapy, and physical therapy are thought to be key regardless of the treatment regimen selected.

From: [1]Department of Pediatrics, University of Wisconsin, Madison, Madison, WI; [2]Department of Anesthesiology, School of Medicine and Public Health, University of Wisconsin, Madison, Madison, WI; [3]Anesthesia and Pain Medicine Service Line, VA Puget Sound Health Care System, Seattle, WA

Corresponding Author: Allison N Heizelman, MD, E-mail: aheizelman@uwhealth.org
Disclaimer: There was no external funding in the preparation of this manuscript.
Conflict of interest: Each author certifies that he or she, or a member of his or her immediate family, has no commercial association (i.e., consultancies, stock ownership, equity interest, patent/licensing arrangements, etc.) that might pose a conflict of interest in connection with the submitted manuscript.
Accepted: 2020-12-08, Published: 2021-06-14

Treatment of refractory ON can be challenging and often requires more aggressive management strategies. While representing an off-label use, recent publications support implanted peripheral nerve stimulator (PNS) treatment in adults (1,5). These devices apply the gate control theory of pain, which describes how the "gate" for transmission of painful stimuli can be halted by providing nonnoxious stimuli (6). PNS devices transmit high-frequency vibrations, halting the transmission of painful input to the central nervous system, thus decreasing perceived pain sensation. We present the case of a 17-year-old boy with a history of significant lesser occipital neuralgia and treatment failures with both medication trials and surgical decompression in conjunction with physical and psychological therapies. After a review of the literature, we were unable to find any reports describing this technique in the pediatric population. Of note, this case report followed CARE Case Report Guidelines (7). Written permission was also obtained prior to submission from the patient and the patient's parent regarding publication and sharing of photos.

CASE

Our patient is a 17-year-old boy with spina bifida and hydrocephalus requiring ventriculoperitoneal (VP) shunt placement, multiple shunt revisions, and a tethered cord release. At age 14, he developed right-sided lesser ON. His pain was 10 of 10 despite medical therapy, including nonsteroidal anti-inflammatory drugs, tricyclic antidepressants, selective serotonin, and norepinephrine reuptake inhibitors, long- and short-acting opioids, benzodiazepines, and gabapentin. Physical therapy and occupational therapy were attempted, although difficult due to the severity of his symptoms. Throughout his treatment, our patient was also connected with psychological therapy. Despite these treatments, his symptoms progressed to extreme allodynia, such that even minor contact with his occiput caused catatonic, unresponsive episodes, requiring him to be strapped to his wheelchair so he did not fall during these spells. He was unable to attend school due to symptoms. He also failed other invasive treatments, including local steroid injections and surgical occipital nerve decompression. We opted against recommending pulsed radiofrequency ablation due to the associated risk of damaging shunt tubing with high temperatures.

He then received a series of biweekly lesser occipital nerve blocks with 0.5% bupivacaine with ultrasound guidance under deep sedation with propofol. Each of the 3 injections provided complete resolution of symptoms, decreasing pain from 10 of 10 to 0 of 10 for a period of 5 to 7 days, demonstrating a pattern of consistent improvement with this intervention. Because of this, we felt he was a candidate for a PNS trial. Since the available device is not currently approved by the US Food and Drug Administration for pediatric patients, there was an unanticipated delay in obtaining approval for the procedure. He required weekly occipital nerve blocks in the interim, which continued to provide complete, temporary relief.

The patient underwent a temporary PNS (StimQ Neurotransmitter Spare Lead Kit, Stimwave© LLC, Pompano Beach, FL) trial, placed under ultrasound guidance with significant improvement. His pain resolved quickly after placement and remained gone for the duration of the trial. Therefore, he opted for placement of a permanent PNS device.

The permanent, wireless PNS (StimQ Neurotransmitter Receiver Kit, Stimwave© LLC, Pompano Beach, FL) was placed percutaneously with ultrasound guidance, with the 4-contact lead successfully tunneled deep to his existing VP shunt (Fig. 1).

During the procedure, the patient received a propofol infusion and intermittent fentanyl boluses while maintaining spontaneous ventilation. Positioning was prone with slight flexion of the cervical spine. The skin was prepped with chlorhexidine and local 2% lidocaine injected prior to incision. A small skin nick was made inferior and slightly lateral to the C3 vertebral body, which is where the device introducer needle was advanced just inferior to the lesser occipital nerve; the octad 4-contact lead was deployed in close proximity to the nerve. The plastic barbs of the device allow for adequate anchoring to surrounding tissue.

After ultrasound confirmation of correct placement, the stylet within the lead was removed and replaced with the receiver. An intraoperative transmitter was then used to verify a proper connection. Sedation was lightened, stimulation with the device initiated, and the patient noted resolution of his symptoms at a frequency of 500 Hz.

After adequate paresthesia was achieved, a 1.5-cm pocket incision was made in the medial aspect of the neck, inferomedial to the VP shunt. The receiver was then tunneled proximally from the subcutaneous pocket to the initial nick site. A second 2-cm incision was made to bring the receiver to the midline of his back. The lead was then sutured within the base of the pocket with 2.0 silk and each of the sites irrigated with antibiotic irrigation. Closure of the wounds was performed with

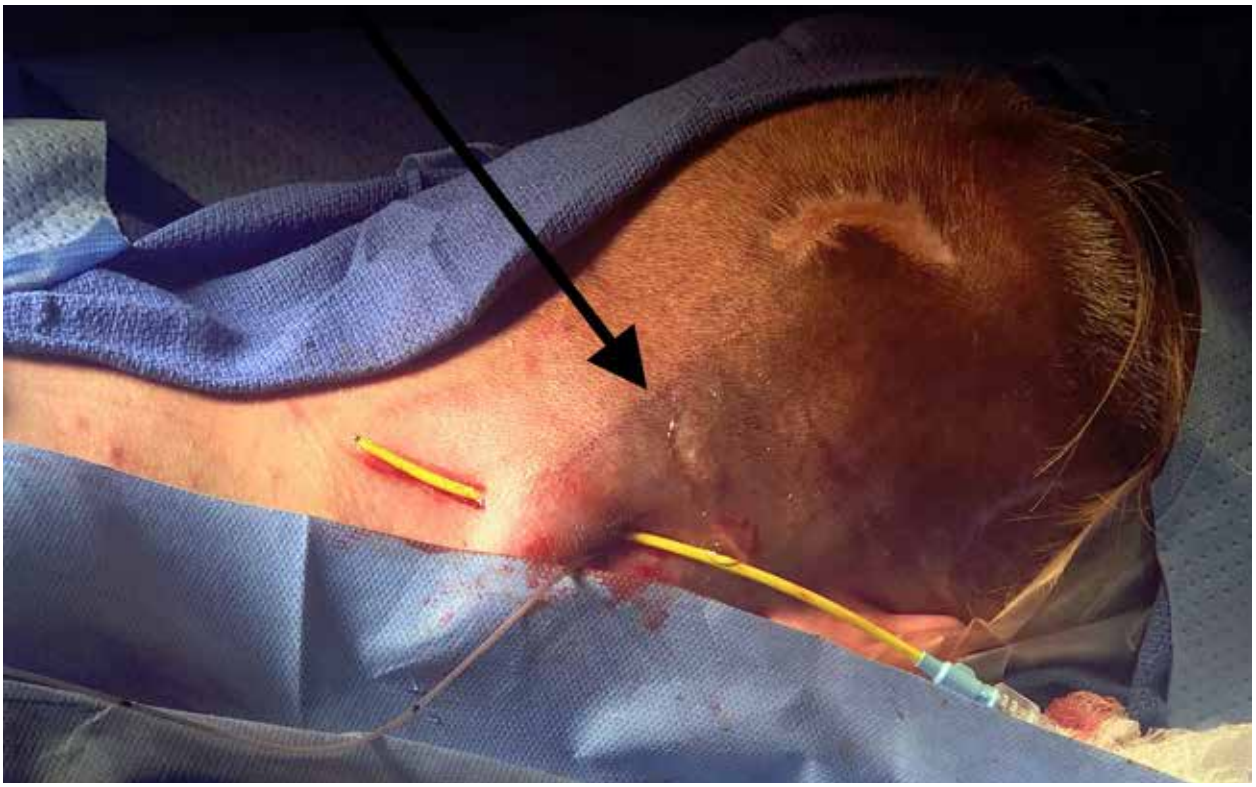

Fig. 1. Tunneling of the permanent peripheral nerve stimulator deep to the existing ventriculoperitoneal shunt (arrow). The scar in the upper right is from a previous operation.

2-0 silk for deep sutures on the larger back site, 4-0 monocryl superficially, and Dermabond®.

There were no complications and the patient experienced immediate relief after placement of the device. At follow-up appointments through 9 months after the procedure, the patient has reported minimal to no ON symptoms and pain scale ratings now 0 of 10 to 4 of 10. He has weaned off amitriptyline, previously prescribed to treat ON symptoms, and resumed physical therapy required for his preexisting spinal issues. There have been no complications related to the PNS. Our patient reports satisfaction with the device, as he was able to resume normal daily activities, including returning to school.

DISCUSSION

To our knowledge, there have been no prior reports describing the use of an implanted PNS device in pediatric patients for the treatment of ON. This procedure is less invasive than other surgical therapies performed for refractory ON and can be a safe and effective treatment option. This procedure also has the potential benefit of reducing or eliminating the use of systemic medications, which can have their own undesirable adverse effects. There is literature demonstrating success in adults with limited complications (5), although the sample sizes are small and long-term data are lacking.

As demonstrated with our patient, the first step towards implantation is to confirm that symptoms involve a nerve pain component; this is done by performing a series of nerve blocks (often at least 2) and confirming significant relief of symptoms with this intervention. Patients should also undergo a psychological evaluation, assessing their understanding of the benefits and limitations of this technology, expectations, and psychological risk factors. It can then be beneficial to do a temporary device trial with percutaneously placed lead(s) to confirm improvement prior to permanent device placement.

A temporary implant was used in our patient. While not necessarily a requirement, there is a small case series documenting a 25% incidence of nonresponders to trial therapy, who therefore did not proceed to permanent

implantation (8). This step appears to aid appropriate selection of those patients who will have relief with the permanently implanted device.

The PNS device use in our patient is wireless without an internally implanted generator, requiring a patient to wear an external battery close to the receiver of the lead. Our patient carries his battery via a small pocket sewn into the neck of his shirts, which has been efficacious. He also has not had any adverse events related to the device; therefore, no events have been reported to the manufacturer or government agencies.

Our case further demonstrates that an occipital PNS can be successfully placed without disruption or damage of an ipsilateral VP shunt. While there is literature suggesting that occipital nerve blocks can be safely administered with a VP shunt (9), we found no such reports for PNS placement. Also of note, pulsed radiofrequency ablation is associated with the risk of damaging the shunt tubing due to the high temperatures used (8), so PNS may be superior to this therapy in this specific population.

PNS placement markedly reduced our patient's ON symptomatology immediately following the procedure and allowed for significant lifestyle improvements long-term. Per CARE guidelines, we include a statement from our patient regarding this: "I was not able to leave the house due to the headache pain, nerve pain, and the dissociative disorder the pain would cause before the stimulator. Since the intervention, I am able to go fishing, hunting, and do many other things with my family and friends that I had not been able to do before. I feel as though I have my life back. One negative is that I am unable to go swimming, as I still need the stimulator on at all times, but I am hoping in the future that I will be able to swim. The only minor inconveniences are that the battery does not tell you when it turns off and does not last very long, but we have figured out how to manage those minor things. I am absolutely satisfied with the nerve stimulator." While noting some minor drawbacks, overall he reports a positive outcome from the PNS implantation with significant improvement in his performance of daily activities at home, in school, and during recreational time.

Lastly, while our teenage patient has physiology similar to an adult, case reports for PNS use in the pediatric population are lacking. Reports documenting the success of these devices in pediatric patients can be the basis for further safety and efficacy trials in younger children. They may also prove valuable when obtaining authorization from institutions and insurances for these procedures in children.

CONCLUSION

Refractory occipital neuralgia symptoms can exhibit dramatic improvement with the percutaneous placement of a permanent PNS device, which is feasible in pediatric patients even with an ipsilateral VP shunt. Further research is warranted to determine PNS efficacy on a broader scale in pediatric patients with refractory neuralgias.

REFERENCES

1. International Neuromodulation Society. About neuromodulation. International Neuromodulation Society Web site. www.neuromodulation.com/about-neuromodulation. Date Updated 02/06/2018. Date Accessed 04/05/2020.
2. Silberstein SD, Dodick DW, Saper J, et al. Safety and efficacy of peripheral nerve stimulation of the occipital nerves for the management of chronic migraine: Results from a randomized, multicenter, double-blinded, controlled study. *Cephalagia* 2012; 32:1165-1179.
3. Headache Classification Committee of the International Headache Society (IHS). The International Classification of Headache Disorders, 3rd edition. *Cephalalgia* 2018; 38:1-211.
4. Barmherzig R, Kingston W. Occipital neuralgia and cervicogenic headache: Diagnosis and management. *Curr Neurol Neurosci Rep* 2019; 19:20.
5. Keifer OP, Diaz A, Campbell M, Bezchlibnyk YB, Boulis NM. Occipital nerve stimulation for the treatment of refractory occipital neuralgia: A case series. *World Neurosurg* 2017; 105:599-604.
6. Mendell LM. Constructing and deconstructing the gate theory of pain. *Pain* 2014; 155:210-216.
7. CARE Case Reporting Guidelines. 2013 CARE Checklist. www.care-statement.org/checklist. Date Updated 11/17/2019. Date Accessed 04/21/2020.
8. Huntoon MA, Burgher AH. Ultrasound-guided permanent implantation of peripheral nerve stimulation (PNS) system for neuropathic pain of the extremities: Original cases and outcomes. *Pain Med* 2009; 10:1369-1377.
9. Kim E, Cucchairo G. Occipital nerve blocks for relief of headaches in patients with ventriculoperitoneal shunts: A case series. *J Child Neurol* 2019; 34:674-678.