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Peripheral Nerve Field Stimulation With Fast-Acting Sub-Perception Therapy (FAST™) in a Patient With Nummular Headache

Sneha C. Balasubramanian, FRCS and Girish V. Vajramani, FRCS

Background: Nummular headache is a rare primary headache disorder that is often difficult to treat and frequently resistant to conventional therapies. We report the first case of a permanent peripheral nerve field stimulation (PNFS) implant utilizing Fast-Acting Sub-perception Therapy (FAST™) for the treatment of nummular headache.

Case Report: A 45-year-old man with a 4-year history of severe right temporal nummular headache was initially successfully managed with percutaneous electrical nerve stimulation therapy. However, lack of long-term pain relief necessitated consideration of a permanent PNFS implant. The procedure involved placement of 2 linear electrodes and an implantable pulse generator. Following activation with FAST™, the patient experienced immediate and sustained pain relief, with resolution of allodynia, and has remained pain-free at 2 years of follow-up.

Conclusions: This case demonstrates the potential efficacy and long-term benefit of peripheral nerve field stimulation using FAST™ therapy in the management of refractory nummular headache.

Key words: Nummular headache; peripheral nerve field stimulation; fast-acting sub-perception therapy; neuromodulation

BACKGROUND

Nummular headache, also referred to as "coin-shaped headache," is a primary headache disorder as per the International Classification of Headache Disorders, 3rd edition and is characterized by intermittent or continuous scalp pain of highly variable duration (1). It is often chronic and in a small, circumscribed area of the scalp no larger than 6 cm in diameter and occurs in the absence of any underlying structural lesion. The pathogenesis is unknown, and the pain can be perceived as a pressure, stabbing or occasionally burning sensation with allodynia. The treatment options include nonsteroidal anti-inflammatory drugs, antiepileptic agents and tricyclic antidepressants. We had earlier described the first successful use of Percutaneous Electrical Nerve Stimulation (PENS) therapy in treating nummular headache (2). This report is of the same patient who was subsequently implanted with a permanent peripheral nerve field stimulator. Peripheral nerve field stimulation (PNFS) is an emerging neuromodulation therapy for the management of refractory neuropathic pain (3-5). We hereby describe the first report of a patient with nummular headache treated successfully with permanent peripheral nerve field stimulator implant with fast-acting sub-perception therapy (FAST™).

From: Centre for Functional Neurosurgery, Hampshire, United Kingdom

Corresponding Author: Girish V. Vajramani, FRCS, E-mail: girish.vajramani@uhs.nhs.uk
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CASE PRESENTATION

A 45-year-old man was referred to Wessex Neurological Centre with a 4-year history of headache restricted to an area, the shape of a coin, in the right temple (Fig. 1). His medical comorbidities included asthma, depression, hypertension, and obesity. A magnetic resonance scan of the head was normal with no evidence of any trigeminal neurovascular conflict. A diagnosis of nummular headache was made based on the classical location and symptoms. His pain ranged from a constant 3/10 to exacerbations up to 10/10 on the numeric rating scale (NRS) with an average pain score of 7/10. This was also associated with allodynia to the cold and to light touch. After failed conservative treatment with neuropathic pain medications, including carbamazepine, pregabalin, and other agents, he was treated with local anesthetic and steroid injections and cryotherapy. The pain relief with the injections and cryotherapy was short lasting and he continued to have severe hypersensitive exacerbations. During the flare-ups, he was unable to touch the temple, wash the area, wear glasses or a hat. He was then managed successfully with PENS therapy. A 20-mm electrode (Algotec Ltd) was used in the region of pain with standard stimulation parameters for 25 minutes (3-second alternating cycles of 100 and 2 Hz) at 2-3 V strength as tolerated. He underwent 6 sessions of PENS therapy over a 4-year period each offering up to 4 months of effective pain relief and decreased sensitivity over the pain area with a noticeable cumulative effect (2). However, he continued to have severe exacerbations and failed to have long-term pain relief.



Fig. 1. Clinical photograph showing the region of pain with dystrophic skin changes.

Following the success of the PENS therapy, the patient was taken up for permanent implantation of a PNFS device as a long-term pain relief option. The procedure was done under general anesthesia; 2 Linear ST percutaneous electrodes (Boston Scientific Ltd, Valencia, CA, USA) were inserted in the right temporal region subcutaneously and electrodes were anchored to the fascia (Fig. 2). The implantable pulse generator was implanted in the right infraclavicular region (Wave Writer Alpha IPG, Boston Scientific Ltd, Valencia, CA, USA). The device was activated immediately after surgery. The patient demonstrated good and immediate pain relief in the region of nummular headache. He also had numbing of the region and decreased sensitivity to touch.

Programming and Outcomes

The device was switched on postoperatively, and the patient exhibited an immediate positive response to stimulation, allowing for precise programming using a combination of FAST™ Therapy and low-frequency tonic stimulation (2–100 Hz) at therapeutic amplitudes. The stimulation-induced paresthesia overlapped the painful area of the nummular headache. Currently, he uses a combination of FAST™ Therapy and paresthesia-based programs. The configured stimulation parameters effectively covered 100% of the patient's painful area. A continuous cycling schedule was established, alternating 6 minutes of tonic stimulation with 6 minutes of FAST™ Therapy. All electrode contacts on the leads were utilized, and no further optimization has been required since the initial programming.

The immediate onset of FAST™ Therapy resulted in rapid and sustained pain relief, with the patient's NRS pain score decreasing from 7/10 preoperatively to 0/10 after device activation. Pain levels ranged from 0/10 to 2/10 at 3-, 6-, 9-, 12-, and 24-month follow-up, with the patient reporting a pain score of 0/10 at the most recent follow-up (Fig. 3). At 24 months, he reported his condition as "very much improved" on the Patient Global Impression of Change scale. The EQ-5D-5L utility index score improved from 0.585 at baseline to 1.00 at the most recent follow-up, with an overall health rating of 90/100 (Fig. 4). He had discontinued all pain medications, and his sleep and mood had substantially improved. At 2 years postoperatively, he was pain-free and described the improvement in his quality of life associated with stimulation as "life-changing."

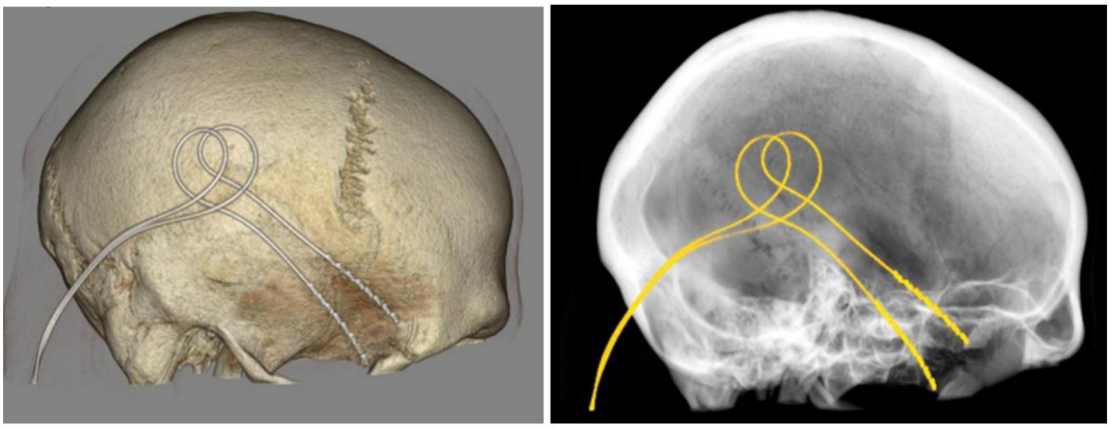


Fig. 2. Computed tomography images (3-dimensional reconstructions) showing placement of the electrode.

DISCUSSION

Nummular headache is an uncommon variety of headache and challenging to treat. The pathophysiology of nummular headache remains uncertain. Some authors believe that it is a condition involving the pericranial soft tissues whereas others believe that it is neuropathic pain with damage to small nerve fibers and sensitization of primary sensory neurons subcutaneously (6). Both central and peripheral mechanisms contribute to the chronic pain often associated with sensory changes overlying the affected region (6). The episodes can become chronic associated with sensitization and exacerbations. While the traditional and conservative medical management options might be successful for the mild to moderate variants, severe and persistent nummular headache is often medication refractory. There are case reports of successful botulinum toxin injections and transcutaneous electrical nerve stimulation therapy. However, the pain relief from these procedures can be short-lived and neuromodulation therapies can potentially offer long-term pain relief.

Invasive neuromodulation for headache and craniofacial pain is well established treatment with varied evidence base (7). The techniques include occipital nerve stimulator, supraorbital and supra trochlear nerve stimulation, peripheral nerve field stimulation, sphenopalatine ganglion block stimulation, spinal cord stimulation, trigeminal stimulation and auriculo temporal nerve stimulation etc (7). Occipital nerve stimulation, labelled variably as a peripheral nerve stimulator (PNS) or a peripheral nerve field stimulator (PNFS), is mainly indicated in intractable migraine, occipital neuralgia and cluster headache (5,7-10). Peripheral nerve field stimulation (PNFS), an emerging area, targets subcutaneous small-fiber nerve endings rather than a single named peripheral nerve and has shown benefit in several small studies of refractory headache and facial

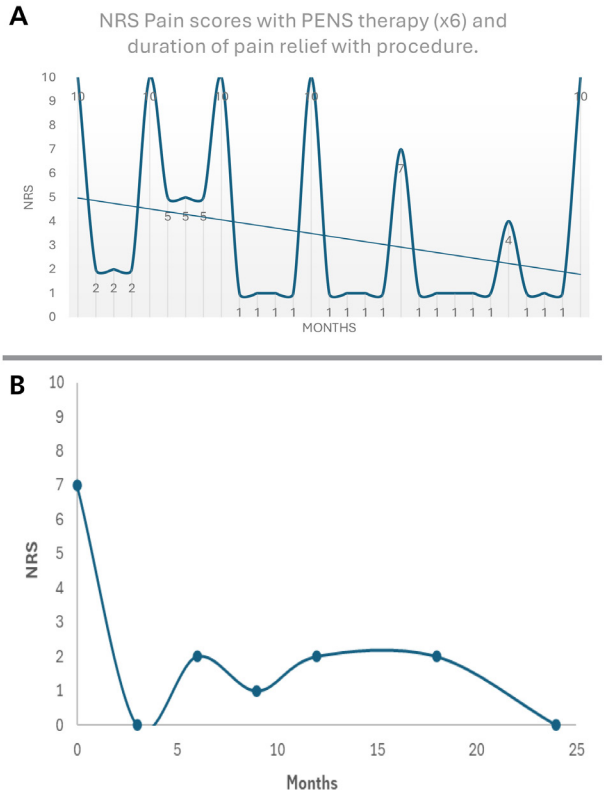


Fig. 3. (A). Graph depicting pain relief with PENS procedures. (B). Graph depicting NRS in the post operative period following PNFS implant.

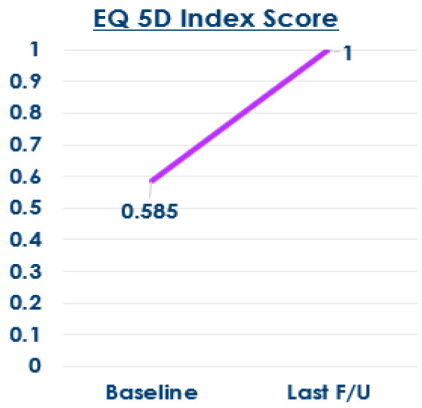


Fig. 4. Graph depicting the EQ-5D index score at baseline and at the 2-year follow-up.

pain (3-5). Cohort and case-series data report substantial improvements, including pain reductions up to ~83%, fewer attack frequencies (~73%), medication reduction in most patients, and some cases achieving complete pain relief after implantation (4,5,7). In a paper by Deogaonkar et al (3), the mechanisms of action of peripheral nerve field stimulation have been postulated as involving the peripheral pathway including conduction block and competitive block of C-fibres and the central pathway via the medial pain pathway modulating the nociceptive processing of pain. Although prospective controlled trials remain limited, existing evidence suggests PNFS is a promising neuromodulation option for treatment-resistant craniofacial pain syndromes.

We hereby describe the first successful use of PNFS with FAST™ therapy to treat nummular headache. We have not come across any previous instances of using FAST™ therapy, although there is one case report in German literature on the use of peripheral nerve field stimulation for nummular headache (11). Our patient responded to the initial PENS treatment, although he did not get long-lasting sustained benefit from this therapy (2). Hence it was decided to convert to a permanent nerve field stimulator implant which has now given him excellent pain relief for almost 2 years. The PENS treatment served as a PNFS trial before the full implant (2).

PENS is a well-established treatment in patients with craniofacial pain syndrome (12). It has low procedure morbidity and can easily be done in outpatient clinic setting. The exact mechanism is uncertain, although alterations neural modulation as well as release of endogenous morphine like substances in the central nervous system may contribute to PENS induced analgesia (12).

A combination of conventional frequency programming and FAST™ Therapy provided significant pain relief in this patient. The mechanism underlying FAST™ Therapy in peripheral nerve field stimulation remains uncertain. FAST™ Therapy has been proposed to preferentially stimulate surrounding fibers while reducing stimulation of central fibers (13). We hypothesize that a similar mechanism may occur with peripheral nerve field stimulation, potentially allowing differential modulation of central and surrounding fibers through the multiple independent current control system. In this patient, such modulation may have contributed to the observed pain relief. To our knowledge, FAST™ Therapy has not previously been reported for peripheral nerve field stimulation.

CONCLUSIONS

Nummular headache is a rare primary headache disorder and neuromodulation therapies are being increasingly used for chronic medication-refractory cases. Our case report is the first to describe the successful use of PNFS with FAST™ therapy in a patient with nummular headache who demonstrated good pain relief with PENS therapy. Results from this patient show significant long-term reduction in pain scores. Although the mechanism of action of FAST™ therapy for treatment of nummular headache is only hypothesized, our results and other case reports suggest that PNFS implant and stimulation therapy is effective. Further studies would be needed to determine if PNFS with FAST™ is the best course of treatment for this uncommon indication, however we feel it can be considered as an option for long-term pain relief in patients with severe nummular headache.

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