

Treatment of chronic intractable atypical facial pain using peripheral subcutaneous field stimulation

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Introduction: Atypical facial pain (ATFP) is challenging to manage and there are few proven therapies available. We present a case report describing application of peripheral subcutaneous field stimulation (PSFS) to a patient with chronic intractable ATFP which conventional treatment failed to ameliorate.

Methods: The patient underwent an uneventful PSFS trial with percutaneous placement of two temporary eight-electrode leads (Medtronic Inc, Minneapolis, MN, USA) placed subdermally over the left mandible.

Results: After experiencing excellent pain relief over the next two days, the patient was implanted with permanent leads and rechargeable generator two and a half weeks later and reported sustained pain relief at 12-month follow-up visit.

Discussion: Peripheral subcutaneous field stimulation provides an effective treatment option for patients suffering from chronic ATFP who have failed conservative treatment. PSFS may provide pain relief with advantages over conservative treatments and more invasive techniques.

Conclusion: Peripheral subcutaneous field stimulation offers an alternative treatment option to select patients with intractable ATFP.

Keywords: Atypical facial pain, electric stimulation, intractable jaw pain, pain, peripheral subcutaneous field stimulation, trigeminal neuralgia

Conflict of interest: The authors reported no conflicts of interest.

INTRODUCTION

Atypical facial pain (ATFP) represents a wide group of facial pain problems which have many different causes but present with common symptoms. Some studies postulate a low-grade infectious and inflammatory process occurring over a long period can result in nerve damage and be the triggering factor for ATFP. Dental or physical trauma also is linked to ATFP. Malignant neoplasms invading the base of the skull and traumatizing branches of the trigeminal nerve and even benign tumor of the trigeminal nerve or meninges can lead to ATFP (1). There was report of facial sarcoidosis presenting as ATFP (2). In some of the patients no etiologic factors can be found and patients in this group tend to have significant psychopathology (3). Successful treatment of the patients with ATFP is rare because of the variety of etiologies of this syndrome.

Atypical facial pain differs from trigeminal neuralgia (TN) in every respect. The ATFP is reported as continuous but can fluctuate in intensity. This pain described as burning, aching or cramping, pinching, pulling, often in the region of trigeminal nerve and can extend into the upper neck or back of scalp. TN is characterized by paroxysms of severe, lancinating, electric-like bouts of pain restricted to the distribution of trigeminal nerve. The pain may last from several seconds to minutes. Attacks are often triggered by eating, brushing teeth, and washing. Between the paroxysms the patients are free of symptoms but report experiencing the fear of an impending attack. Sufferers go out of their way to avoid any contact to trigger areas (4).

This disorder of the sensory division of the trigeminal nerve may be due to degenerative, fibrotic changes in the gasserian ganglion or due to tortuous blood vessels compressing the trigeminal root as it exits the brain stem, as can occur in the patients with MS. Other causes of TN include tumor growth, bony abnormalities, and other vascular conditions. Interventional treatments included in the management algorithm for patients with TN include microvascular decompression (MVD) and percutaneous stereotactic radiofrequency (PSR) rhizotomy. MVD and PSR rhizotomy have comparable rates of pain relief that are highest among available options and can approach 77% in seven years for MVD and 75% in six years for PSR rhizotomy (5).

Traditional treatment options for ATFP include anticonvulsants, antidepressants, baclofen, triptans, non-steroidal anti-inflammatory medications, and opioids. ATFP is challenging to manage and there are few proven therapies available. Treatment

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with these medications is often ineffective and wrought with intolerable side-effects. Surgical procedures such as MVD generally are not successful with ATRP patients. Peripheral nerve stimulation has been used to treat a variety of neuropathies (6), including ilioinguinal (7), occipital (8–11), post-herpetic (12), and stimulation of trigeminal branches was reported for treatment of trigeminal postherpetic neuralgia and trigeminal post-traumatic neuropathic pain (13–16) with excellent relief of pain. Treatment of ATRP, including trigeminal neuropathic pain using peripheral subcutaneous field stimulation (PSFS) can lead to decreased reports of pain as well as a reduced need for oral pain medications.

CASE REPORT

A 72-year-old woman was referred to our clinic with a history of left jaw pain, previously diagnosed as TN. Her pain had begun more than three years ago. She was followed by an oral surgeon and orthodontist for jaw pain and temporomandibular joint disorder. Tooth extractions and trigger point injections inside the left lower gum were unsuccessful at treating her pain. She had been seen by a pain physician two years before treatment in our clinic who had performed a trigeminal nerve block which provided several hours of relief. Subsequently, the patient underwent stereotactic radiofrequency ablation of the gasserian trigeminal ganglion for the second and third trigeminal nerve division. The patient received no relief from this procedure. The patient was evaluated by neurosurgeons and was not considered to be a candidate for gamma-knife radiosurgery and MVD.

Previous conservative therapy had included trileptal, pregabalin, darvocet, oxycodone, fentanyl patch, gabapentin, nonsteroidal anti-inflammatory medications, and topical ointments, none of which provided the patient relief. Upon presentation to our clinic the patient's chronic pain medication regimen included long and short acting oxycodone and ibuprofen. The patient described her pain as aching, burning, rarely sharp in character and lasting all day and night. The pain could get better with sleep. Distribution of the pain was over left mandible and mastoid process. On physical examination we found allodynia over the left proximal mandible; multiple teeth were removed on the left. In our opinion the patient had a clinical presentation of ATRP but not a classic TN. A trigeminal nerve block performed in our clinic provided three weeks of pain relief. The patient was counseled on treatment options including continued treatment with oral pain medications, or peripheral nerve stimulator therapy. The patient elected to proceed with peripheral nerve field stimulator therapy.

The patient underwent successful two-day trial of percutaneous placement of two eight-electrode leads (Medtronic Inc., Minneapolis, MN, USA) after passing a psychological evaluation for an implantable device. Leads were placed subdermally over the left mandible (Fig. 1). During the PSFS trial, she reported greater than 50% improvement in pain and rated her pain as a 2 on the visual analog scale (VAS) compared with a 9 on the VAS before trial leads were placed. Two and a half weeks later the patient underwent implantation with permanent leads and a RestoreUltra (Medtronic Inc., Minneapolis, MN, USA) rechargeable generator. Preoperatively we discussed with the patient location of the generator and she chose supragluteal area because of cosmetic concerns. The patient had a previously placed Port-a-catheter for chemotherapy and did not want new scars over the chest. The procedure was performed in an ambulatory surgery center with intravenous sedation and local anesthesia administered by the surgeon. We chose right lateral



Figure 1. Subdermal placement of two eight-electrode leads showing the electrodes placed over the left mandible.

decubitus position for easy access to left side of the face, neck, and back. Two permanent eight-electrode standard Octad leads were inserted subdermally along left mandible through vertical 1.5 cm incision 2 cm anterior to the tragus of the left ear. The leads were passed through slightly bent 14 Gauge Tuohy needles to follow the curvature of the mandible. Both leads were anchored in the wound to fibroaponeurotic tissue with 2-0 nonabsorbable suture of braided polyester (Ethibond) and Titan anchors (Medtronic Inc., Minneapolis, MN, USA). The leads were tunneled over and behind the ear to the second incision created over the upper posterior neck where they were connected to extensions. Extensions were finally brought by use of a 60-cm tunneling tool into the left supragluteal area to the subcutaneous pocket created for the generator and were connected to a RestoreUltra (Medtronic Inc., Minneapolis, MN, USA) rechargeable generator. The post-operative course was uneventful. Initiation of use of RestoreUltra rechargeable generator was uneventful during the post-operative period.

The stimulator was programmed using a guarded electrode configuration with a pulse width of 450 msec and a rate of 60 Hz. The amplitude use ranged from 1.5 to 2.3 V. The patient reported that the stimulation covered 100% of her painful areas following the initial programming. After implant surgery the patient was weaned off all opioids. The patient has been using her PSFS 24 hours per day, adjusting stimulation intensity for changes in intensity of pain with good pain relief. She continued to report excellent pain relief at her 12-month follow-up visit.

DISCUSSION

The PSFS alleviates pain by subdermal stimulation of the peripheral fibers, which may prevent transmission of painful impulses to the central nervous system. The neuromodulating effects of electrical stimulation are based on the tenets of the "gate-control theory" of pain proposed by Melzack and Wall in 1965 (17). Based on this theory, it is hypothesized that PSFS "closes the gate" to pain transmission by activating large-diameter afferent fibers via application of an electric field. PSFS may also alter local blood flow, cause release of endorphins, affect neurotransmitters and axonal conduction, and may block cell membrane depolarization (18). The mechanism of action of PSFS and neuromodulation in general continues to be investigated as there may be a multitude of ways in which neuromodulation affects pain transmission. PSFS can be effective in treating painful areas, such as the face, which are very difficult to target with epidural stimulation.

Peripheral subcutaneous field stimulation is an alternative treatment option for patients suffering from chronic AFTP. PSFS has many advantages over many conservative treatments as well as more invasive techniques. There are no side-effects created by PSFS as there are with many medications. There is a high rate of success with permanent implant due to the fact that a trial is performed during which the patient evaluates the efficacy of the device. The therapy is completely reversible if for some reason therapy becomes contraindicated or is no longer needed. Patient programmers permit patients to control the level of stimulation they feel based on their degree of pain. This enables patients to take a more active role in their pain management.

SUMMARY

We present a single case of intractable AFTP which was refractory to conventional treatment but successfully treated with PSFS. This technique may be a safe and effective treatment for patients who have failed to find relief with more conservative measures or who are not appropriate candidates for more invasive interventional pain or surgical procedures based on their comorbid health conditions. PSFS has provided our patient with satisfactory pain relief without the side-effects of previous medication therapy. In our opinion, PSFS offers a safe and effective treatment method that is completely reversible should a patient lose its pain-alleviating effect. This case study provides support for PSFS as an alternative treatment option for patients with intractable jaw pain and will inspire interest in prospective studies comparing peripheral nerve stimulation with other therapies.

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COMMENTS

This case report describes use of peripherally applied neurostimulation in the treatment of patient with chronic and severe pain in the lower part of her face. The painful condition is probably best referred to as a trigeminal neuropathic pain, and the authors may be commended for their decision to use this novel modality in this particular location and condition.

In contrast with trigeminal neuralgia, that is successfully treated with variety of surgical interventions (and which would have worsened from electrical neurostimulation), this neuropathic pain is hard to treat with conventional surgical approaches. Therefore, we and others have used peripheral neurostimulation for this patient category (1–6)—but traditionally stayed away from mandibular area due to its high mobility and associated fear of electrode migration and/or fracture. Also, since mandibular nerve is harder to reach (comparing to subcutaneous course of infraorbital and supraorbital nerves), the pain affecting this part of the face has not been treated with true peripheral nerve stimulation—and therefore using peripheral subcutaneous field stimulation may be the only way to provide neurostimulation in this area.

Interestingly enough, although first well-documented mention of peripheral nerve stimulation for pain control involved infraorbital region—when Drs. White and Sweet stimulated their own infraorbital nerves to illustrate pain suppression based on “gate-control” theory (7)—this was done for research purposes only. The first clinical use of peripheral nerve stimulation concept in treatment of facial pain was specifically related to a similar neuropathic pain in mandibular distribution back in mid-1960-s (8), although in that case stimulation was delivered at 14000 Hz frequency, something that has not been much explored at a later time.

Therefore, I want to congratulate the authors of this report for continuing this historical tradition and getting good results in a patient who most likely would have been told by many others that there is nothing that may be offered.

I fully agree that more cases like this will be needed before this modality may be recommended for widespread acceptance. In general, peripheral neurostimulation is now enjoying rapid spread of new indications—and among the latest condition for which it has been used are fibromyalgia, back and neck pain, migraine, inguinal pain, etc. The list will likely continue to grow, and it would be very important to document all new peripheral neurostimulation applications and continue diligent follow up of outcomes, side effects and complications.

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This article shows still another application of the new and growing field of Peripheral Subcutaneous Field Stimulation. Neurostimulation options, until now, included motor cortex stimulation or deep brain

stimulation. Both modalities are highly invasive and complex. The described modality is much simpler and less invasive. If its effectiveness is confirmed by a larger clinical series, this could become the first line of approach in the neuromodulation algorithm.

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