

Case Report



Sphenopalatine Ganglion Electrical Nerve Stimulation Implant for Intractable Facial Pain

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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.

Manuscript received:
10-04-2014
Revised manuscript received:
01-05-15
Accepted for publication:
02-20-2015

Free full manuscript:
www.painphysicianjournal.com

Persistent idiopathic facial pain can be extremely difficult and significantly challenging to manage for the patient and the clinician. Pharmacological treatment of these painful conditions is not always successful.

It has been suggested that the autonomic reflex plays an important role in the pathophysiology of headaches and facial neuralgia. The key structure in the expression of cranial autonomic symptoms is the sphenopalatine ganglion (SPG), also known as the pterygopalatine ganglion. The role of the SPG in the pathophysiology of headaches and facial pain has become clearer in the past decade.

In this case report, we describe a 30-year-old woman with insidious onset of right facial pain. She was suffering from daily pain for more than 9 years prior to her visit at the pain clinic. Her pain was constant with episodic aggravation without a predisposing trigger factor. The patient was evaluated by multiple specialties and tried multimodal therapy, which included antiepileptic medications, with minimal pain relief. A SPG block using short-acting local anesthetic provided significant temporary pain relief. The second and third attempt of SPG block using different local anesthetic medications demonstrated the same responses.

After a thorough psychological assessment and ruling out the presence of a correctable cause for the pain, we decided to proceed with SPG electrical neuromodulation. The patient reported significant pain relief during the electrical nerve stimulation trial. The patient underwent a permanent implant of the neurostimulation electrode in the SPG region. The patient was successfully taken off opioid medication and her pain was dramatically responsive during a 6-month follow-up visit. In this article we describe the SPG nerve stimulation and the technical aspect of pterygopalatine fossa electrode placement.

The pterygopalatine fossa is an easily accessible location. This case report will be encouraging for physicians treating intractable facial pain by demonstrating a novel therapeutic option. This report shows a minimally invasive approach to the SPG.

Key words: Sphenopalatine ganglion, persistent idiopathic facial pain, electrical neuromodulation

Pain Physician 2015; 18:E403-E409

The diagnosis and management of patients with persistent idiopathic facial pain can prove daunting even to experienced clinicians. The causes of chronic facial pain are numerous, and misdiagnosis and mismanagement are common.

Sphenopalatine ganglion neuralgia, atypical facial pain, atypical facial neuralgia, and chronic idiopathic facial pain are being used in the literature to describe a

constellation of pain symptomatology in the facial distribution. The management strategies often times do not generate an effective clinical analgesic response. Persistent idiopathic facial pain is a dilemma for the physician and the patients.

Ever since that clinicians and researchers started using the criteria of The International Classification of Headache Disorders, 3rd edition (ICHD-3 beta version),

for headache and face pain there is collectively some consensus on naming the situation "Persistent Idiopathic Facial Pain." The classification is based on the best current knowledge but remains hierarchical. The incidence of persistent idiopathic facial pain is 1/100 000; both sexes are affected equally, but more women than men seek medical care (1). Wide disagreement exists as to the pathogenesis of chronic facial pain syndrome. There is some speculation that the pain is purely due to parasympathetic dysfunction that originates from the ganglion or from more complex central dysfunction.

The key structure in the expression of cranial autonomic symptoms is the sphenopalatine ganglion (SPG), also known as the pterygopalatine ganglion. The SPG lies in the pterygopalatine fossa. The pterygopalatine fossa is a pyramidal space situated behind the posterior wall of the maxillary sinus, anterior to the medial plate of the pterygoid process, and lateral to the perpendicular plate of the palatine bone. The pterygopalatine fossa is limited by the sphenoid superiorly and laterally it communicates with the infratemporal fossa. The pterygopalatine fossa also contains the internal maxillary artery and several nerves including the maxillary nerve, mandibular nerve, the greater and lesser petrosal nerves, and the SPG with its afferent and efferent branches. Sympathetic and parasympathetic fibers join the SPG; the parasympathetic fibers originating from it are distributed to the nasal mucosa and the lacrimal glands (2). The SPG may play a key role in the genesis of facial pain.

Afferent trigeminal and facial fibers from the throat, palate, and nose traverse the SPG before joining the maxillary nerve. Sympathetic efferent fibers transit the SPG en route to their target organs, such as the pupils or sweat glands. Parasympathetic efferent fibers from the superior salivary nucleus synapse in the SPG, and then postganglionic parasympathetic proceed to the end organs, such as pupils, sweat glands, lacrimal glands, and parotids. Therefore the SPG is unique because of its neuronal circuitry and its relationship to the maxillary branch (V2) of the trigeminal nerve (V). The seventh cranial nerve uses the fifth cranial nerve as a pathway or structural vehicle as a passageway for its postganglionic parasympathetic fibers.

Based on this anatomical distribution, we may hypothesize that the centrally activated signals may generate a neurogenic inflammation, which in turn sensitize peripheral nociceptors in the facial distribution, and conversely carry the pain signals centrally. In other words, we may generate a theory that the SPG is the

potential pain generator. The SPG may be a relaying station to carry pain signals from the periphery to the central nervous system.

There is growing evidence of benefits from electrical neuromodulation for patients with neuropathic facial and headache pain of various etiologies. The commonly targeted nerves like the infraorbital, supraorbital, occipital, and, more recently, the great auricular are well described in the literature (3,4).

Based on the same anatomical evidence and clinical findings, we used electrical stimulation of the SPG to did interfere with parasympathetic postganglionic outflow, resulting in termination of neuropathic pain.

There is no similar clinical report in the medical literature about SPG electrical nerve stimulation for chronic idiopathic facial pain management.

CASE DESCRIPTION

The patient is 36 year-old woman, who was referred to the center of pain medicine. She had no prior significant medical history and reported a more than 9 year history of constant daily right face pain with significant episodic attacks.

The patient reported that the pain started insidiously and gradually became daily pain.

The pain characteristics were variable for many years; however, they were mostly reported as lancinating, sharp, shooting, and with deep pain sensation. The pain focused around the right maxillary, infraorbital region with some extension to the mandibular region.

She reported that her pain was aggravating with talking, washing her face, moving her tongue, brushing her teeth, or opening her jaw. The patient occasionally has more pain while swallowing solid food.

The patient has never experienced aura, nausea or vomiting, and auditory or visual problems like hearing difficulties or loss of balance.

Magnetic resonance imaging of the brain, which focused on the posterior fossa, and computed tomography of the temporo-mandibular joint did not reveal neurovascular compression, cerebellopontine angle lesion, demyelinating disease, or any abnormality in the pterygopalatine fossa or temporo-mandibular joints.

Furthermore, nasal endoscopy did not demonstrate any abnormalities. She had a normal electroencephalogram and visual evoked potential.

She had experienced the maximum tolerable dose of antiepileptic medications like carbamazepine, oxcarbazepine, gabapentin, pregabalin, topiramate, and sodium valproate in the past. Antidepressant medica-

tions like amitriptyline provided sedation and minimum pain relief.

The patient stated that she is chronically constipated because of opioid medications. At the time of her first visit with the pain clinic, her daily medication list included gabapentin 3600 mg, clonazepam 1 mg, hydromorphone 12 mg, carbamazepine 400 mg, and citalopram 20 mg.

She denied vision problems. Her gait and balance were normal. She rated her pain between 9 and 10 on the Numerical Pain Rating scale (NPR) during the day and between 7 and 8 out of 10 on the NPR during rest. The patient reported that her pain gradually worsened irrespective of her current medical treatment strategy. There was no history of diabetes, trauma, or infection with this patient.

A trial of masticator muscle trigger point injection generated no significant pain relief. We decided to perform a diagnostic nerve block targeting different nerves. A trial of infraorbital, maxillary, and mandibular nerve blocks failed to provide significant pain relief.

Upon review of the medical documents of the patient and the clinical presentation, we concluded the diagnosis of chronic intractable idiopathic facial pain. The SPG block provided short-term pain reduction. We observed the pain reduction benefit of the SPG local anesthetic blocks during multiple occasions. In order to modulate the pain perception, we decided to proceed with the electrical nerve stimulation trial of the SPG.

In the following section, we will describe the nerve stimulator procedure during the trial and during the permanent implantation.

Sphenopalatine Ganglion Nerve Stimulation Procedure Note

Procedure description during the trial

The trial was done under local anesthetic skin infiltration in a sterile condition with the usual preparation and draping technique. The stimulator lead insertion in the pterygopalatine fossa was done in a controlled step-wise fashion under anterior-posterior and lateral fluoroscopy views (bi-planar view). The needle entry was selected based on surface anatomy landmark and fluoroscopic guidance. While we palpated the mandibular notch, the patient was asked to open and close her mouth gently to make the notch even more prominent. Then the proposed anatomical surface landmark was confirmed with the lateral fluoroscopy view to identify the entry point. We selected the entry point about one

centimeter in front of the tragus in order to prevent facial nerve injury. We followed a linear line from the skin to the pterygo-maxillary fissure by inserting a 14 gauge Tuohy needle.

The bi-planar x-ray was extremely useful for gradual needle advancement until the needle tip reached the final destination, where the tip of the Touhy needle reached the beginning of the pterygopalatine fossa.

At this stage, we chose a 1x8 cylindrical percutaneous lead (1x8 Model 3378 Medtronic Co.). We noticed that the 1x8 compact leads provided enough length in the pterygopalatine fossa to cover the potential anatomical location. The lead insertion was done under live bi-planar fluoroscopy after a negative needle aspiration. The lead advancement was done slowly and gently. The patient tolerated the entire procedure very well. The procedure for the trial placement was done with no sedation. We were able to check the final preferred electrode location, both anatomically with bi-planar fluoroscopy and electrically by using a nerve stimulator electrode.

The nerve stimulator parameters were kept in the small window that did not provide motor stimulation. The parameters provided tolerable sensory stimulation (paresthesia) on the distribution of base of the nose to the upper lips. The nerve stimulator parameters of 0.5 millivolt, 250 Pulse Width, and 40 Hertz provided great pain reduction to 1 – 2 on the NPR during the trial on the fluoroscopy table. We secured the catheter in place with sterile adhesive strips to avoid surgical suturing. The catheter was passed over the ear lobe to the front of the neck. The adhesive tape was applied in multiple locations along the course of the catheter to prevent any accidental dislodgment.

She was able to have a solid night's sleep with no pain. The patient was able to reduce her opioid medication consumption during the trial as well. The patient experienced a remarkably significant pain reduction during 5 days of the trial.

The following section describes the entire procedure for the lead insertion in the pterygopalatine fossa during the permanent implant.

Procedure description during the permanent neurostimulation implant

During the permanent implant, the patient underwent the general anesthesia in the operating room. The patient was placed supine on the operating table with her head turned to the opposite side on the horse-shoe radiolucent headrest (Fig. 1).



Fig. 1. This picture shows the patient's position, intraoperative facial nerve monitoring electrode on the frontal muscle (NIM-Response 3.0; Medtronic Co.), marked skin on temporalis muscle (location for anchor device), and marked infraclavicular region (for implanted pulse generator pocketing).



Fig. 2. Postoperative anterior posterior x-ray view.

The pterygo-maxillary fissure was localized using the lateral fluoroscopic view. We followed a linear line from the skin to the pterygo-maxillary fissure by inserting 14 gauge Tuohy needle. The needle was inserted in the infrazygomatic fossa and above the mandibular incisure. The correct lead position was repetitively checked with anterior-posterior and lateral fluoroscopic (bi-planar) views. The tip of the Touhy needle reached at the beginning of the pterygopalatine fossa entrance, then a nerve stimulator lead (Compact Cylindrical Percutaneous Leads 1x8 Model 3378 Medtronic Co.) was gradually inserted and advanced under fluoroscopic bi-planar guidance until it reached to the final position (Fig. 2 and Fig. 3).

During the permanent implant we did not do the electrophysiological testing. However, we were able to obtain the electrical responses in the recovery room in the same fashion as described in the trial lead placement.

The electrode was sutured to the skin with plastic anchors and fine nylon around the insertion site and above the masseter muscle with great attention as to not violate the visible facial nerve branches. The lead tunneled to the right side of the temporalis muscle where we placed the first strain-relief loop under the temporalis fascia to avoid inadvertent electrode pullout. The electrode lead was tunneled toward the generator



Fig. 3 Postoperative lateral x-ray view, showing the lead position and anchor device on temporal region.

pocket. Location of this pocket was chosen based on the patient's and surgeon's preference. In this case the generator was implanted into the right infraclavicular area (Fig. 4).

In the permanent implant, we exclusively relied on the anatomical electrode positioning. The anatomical localization has high reliability of appropriate coverage due to the fact that the ganglion is located in a relatively small anatomical box. Also, the lead tunneling along a relatively long distance is quite painful and necessitates the use of general anesthesia.

The initial programming was performed in the recovery room to produce adequate paresthesia in the nasal-labial area. The patient reported great pain reduction during 6 months follow-up after a permanent neurostimulator implant. She reported significant pain reduction on chronic daily pain with an average of 2 out of 10 on the NPR scale. She was able to completely wean off opioid medications. The patient demonstrated a steady pain reduction with the nerve stimulator parameter choices of 0.5 millivolt, 250 – 450 Pulse Width, and 40 – 80 Hertz.

Discussion

Chronic facial pain, like all chronic syndromes, poses a major challenge to medicine. They are often associated with significant distress, disability, and expenditure of medical resources.

The causes of chronic facial pain are numerous (Table 1).

Detailed pain history and physical examination with proper radiological investigation can eliminate the treatable causes for chronic facial pain.

On the other hand, an extensive investigation can lead to patients feeling ill-understood, over-investigated, and dissatisfied.

The disorder descriptions and treatment tend to be influenced by the background of the specialist assessing the patient. The treatment offered to a patient with chronic facial pain should be addressed with a multidisciplinary approach. Chronic facial pain is a more descriptive term. Although there is an obvious need for prompt elimination of possible organic disease, there is also the danger of overzealous investigation and lack of aggressive multimodality management in the early stage. Multimodality and multidisciplinary approaches should be adopted for diagnostic and prognostic assessment.

There is no treatable cause for a significant number of patients with chronic facial pain. Thereafter, the

Table 1. *Chronic facial pain correctable and non-correctable causes.*

Correctable causes:
Dental Causes
Maxillary sinus
Salivary gland: tumor, blockage, infection, stone
Temporomandibular Joint Disorder
Non -correctable causes:
Idiopathic: burning mouth syndrome
Psychological
Factitious
Fibromyalgia
Neuralgias, Painful Cranial Neuropathies
Trigeminal neuralgia
Trigeminal Neuropathies including deafferentation pain: Post herpetic neuralgia, post traumatic, post surgical, tumor, Multiple Sclerosis, Post Stroke,
Trigeminal Autonomic Cephalgias: short unilateral neuralgiform pain with conjunctival injection, tearing, and redness (SUNCT); and short unilateral neuralgiform pain with cranial autonomic features (SUNA)
Persistent Idiopathic Facial Pain PIFP (Atypical Facial Pain)
Autoimmune: Giant Cell Arteritis
Other (continuous or episodic facial pain): Cluster Syndrome, nervous intermedius neuralgia, Glossopharyngeal Neuralgia, migraines, Tension Headaches, Medication Overuse Headache, ocular myositis, Optic Neuritis, Tolosa- Hunt Syndrome, Raeder Syndrome, Recurrent Painful Ophthalmoplegic Neuropathy, Superior Laryngeal neuralgia, Carotidynia,

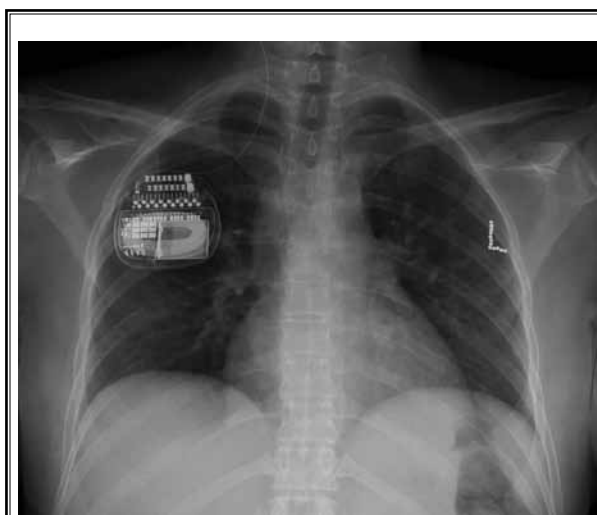


Fig. 4. *The implanted pulse generator location.*

mainstays of treatment are managing the pain, not treating a specific condition, and psychological counseling with combined medication management including antiepileptic and antidepressant medication. Most medical practitioners are often reluctant to prescribe antidepressant and antiepileptic agents for chronic facial pain. In addition, inadequate dosage and duration, perhaps in combination with expressed lack of confidence in their efficacy, prevent the drugs from having any beneficial effect. It is crucial that physicians and patients reach an agreement about how to manage the chronic facial pain.

The SPG has been a target for neuromodulation owing to its links to the trigeminovascular system. Ansarinia et al (5) investigated SPG stimulation in 6 cluster headache patients and found that complete resolution of pain was seen within 3 minutes in 11 out of 18 cluster headache attacks treated.

Similarly, in another institution, a novel device was implanted at the pterygopalatine fossa in 28 patients who randomly received full, sham, or sub-perception stimulation for cluster headache (6). In this case report, we described a unique case of chronic idiopathic facial pain. We utilized the same percutaneous electrode lead technology.

In addition to fluoroscopic guidance on the anterior-posterior and lateral view, the location of paresthesia plays a significantly major role in obtaining further physiologic guidance as to whether the lead is in the proper location to the SPG stimulation. We noticed that paresthesia in the back of the nose and back of soft palate were associated with the expected clinical response due to the proximity of the stimulator lead to the SPG and stimulation of posterior nasal branches. If paresthesia are involving the teeth, gums, or mid or front of palate, this would suggest alveolar or palatine nerves stimulation, respectively, therefore it may not be well accepted by the patient, even though pain reduction may still be achievable.

In this case we were able to obtain a great anatomic and physiologic localization. The pterygopalatine fossa is relatively a small area. One single lead (compact electrode 4 or 8 contacts) can be easily distributed along the anatomical area. The electrophysiological response to the stimulator is easily achievable as long as this ganglion potential anatomic location happens to be adjacent to one of the electrode's contacts, or between 2 contacts. This length distribution of 4 or 8 contact leads will give the opportunity to eliminate some of the electrodes that are not adjacent to the SPG. This

will provide other contacts available for utilization if lead migration happens.

The 4 to 8 contacts leads will enhance the ability to ensure proper stimulation of the SPG over other structures such as the maxillary or palatine nerves. The potential pitfalls with this procedure may include injury to deep vessels, facial nerve, and parotid, as well as piercing bony structures.

In recent years, the blockade of the SPG has increased substantially in interventional pain management for the treatment of refractory headache and facial pain. In order to obtain a longer duration of pain relief benefit, modalities like SPG radiofrequency in the medical literature reported variable clinical responses. One very important reason for SPG radiofrequency failure is the individual variation of the SPG anatomical location (7).

A noninvasive approach, like stereotactic radiosurgery, relies completely on the exquisite visualization of the SPG with modern imaging and our understanding of the anatomy of this region. It is possible that stereotactic radiosurgery of the SPG will become an important option for the neurosurgical treatment of the craniofacial pain. There is no consensus upon using stereotactic radiation as the modality of choice at this time.

The long-term efficacy of deep brain stimulation and motor cortex stimulation for highly selective neuropathic facial pain etiologies are also described in the literature (8,9).

CONCLUSION

The facial pain classification is not concise and complete. There are several treatment strategies available in the medical literature for chronic facial pain. Some physicians who have an accumulation of knowledge and experience in certain applications have been naturally striving to treat every type of facial pain with only one technique of treatment. In chronic under-treated patients, we may see a clinical symptomatology of peripheral sensitization along with more central sensitization. These combined neural sensitizations can become significant obstacles for all the treatment modalities success.

We suggest that multidisciplinary and multimodality treatment strategies be implemented from the early stage of a patient's encounter. It is quite clear that each type of facial pain may require a patient-specific evaluation and different techniques of treatment. The SPG region is a crossroad of sensory, sympathetic, and parasympathetic fibers that when dysfunctional can cause

severe and variable symptoms involving the face. This case report suggests a remarkable neuromodulatory role for SPG electrical stimulation in the treatment of medically refractory facial pain.

As the experience with the use of peripheral nerve stimulation for craniofacial pain treatment grows, one may expect a better definition of the criteria that's predictive of a lasting beneficial outcome. Publication of a large clinical series will likely result in the acceptance of this treatment approach; its minimally invasiveness, testability, reversible effects, and adjustable settings may make it a preferred modality for otherwise intractable conditions. More understanding of the craniofacial pain anatomy and pathology along with a better recognition of the SPG role will improve the strategy and the concept. There is a clear need for optimal lead placement over the SPG within the sphenopalatine fossa.

Author Contributions

Drs. Foad Elahi MD and Dr. Chandan G. Reddy had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. Drs. Elahi and Reddy designed the case study and managed the literature searches and summaries of previous related work and wrote the manuscript.

Conflict of Interest:

Drs. Elahi and Reddy have no conflicts of interest to report. Both authors of the manuscript received no remuneration, no reimbursement or honorarium in any other manner.

The authors are not affiliated in any manner with any company.

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