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Peripheral nerve field stimulation for otalgia: A novel therapy for refractory deep ear pain

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Abstract

Refractory otalgia or deep ear pain is a complex clinical problem that poses significant challenges to the physicians. Here we report a case of a 39 year old female who presented to us with deep right ear pain which started following cholesteatoma excision 11 years ago. Since onset of right ear pain, she had multiple ear surgeries including microvascular decompression and excision of right temporal bone before presentation. Following neuropsychological assessment and excluding underlying depression/anxiety, she underwent peripheral nerve field stimulation (PFNS) trial. She had a successful PFNS trial and underwent permanent implantation of PFNS and pulse generator. She had >50% reduction in her pain intensity on VAS and pain medications. She required explantation due to superficial infection; however she was satisfied with her therapy and looking forward for reimplantation. We report the first case of successful management of refractory deep ear pain using PFNS with a review of pertinent literature.

Introduction

Otalgia or deep ear pain is a complex clinical problem that poses significant challenges to the physicians in terms of both differential diagnosis and management. Neuralgic deep ear pain can be associated with either glossopharyngeal neuralgia or nervus intermedius (geniculate neuralgia) neuralgia as one of the symptoms [1,2]. According to the International Classification of Headache Disorders (2nd edition), presence of a trigger factor is mandatory to establish the diagnosis of either glossopharyngeal or nervus intermedius neuralgia [3]. However there are reports of neuralgic otalgia in the absence of obvious trigger factors [4].

Both glossopharyngeal and geniculate neuralgias are rare disorders with an incidence of 0.2%–1.3% and only 150 reported cases so far in the literature respectively [1,2]. The common surgical options are either microvascular decompression of the nerve at the nerve root entry zone or sectioning of the nerve. However there remains a cohort of patients who are not relieved of their pain following these surgical procedures [5–7]. There has been a burgeoning interest in the utility of peripheral nerve field stimulation (PFNS) for complex cranio-facial pain and other regional pain syndromes [8–10]. This modality has not yet been explored in the management of patients with refractory otalgia. Here we report the first case of successful management of refractory deep ear pain using PFNS with a review of pertinent literature.

Case illustration

A 39 year old married Caucasian female presented to our office with a long history of right deep ear pain. Her right ear pain started following cholesteatoma (benign lesion consisting of keratinizing squamous epithelium and erodes the mastoid process of temporal bone and skull base) excision and insertion of tube in her ear 11 years ago (Fig. 1 a & b). Since then she had multiple right ear surgeries including meataloplasty, revision canaloplasty, tympanoplasty, mastoidectomy, insertion of multiple ear tubes. In addition, she underwent repair of her right tympanic membrane, removal of bony spurs and excision of cholesteatoma. She also underwent sectioning of her right vestibular nerve due to persistent pain. She was diagnosed to have right glossopharyngeal neuralgia and underwent microvascular decompression of right glossopharyngeal nerve 4 years ago and the postoperative course was complicated with CSF leak. This surgery was also met with unsatisfactory results. A year later she underwent tympanoplasty without ossicular chain reconstruction and ultimately had resection of her right temporal bone 2 years prior to presentation due to persistent right deep ear pain (Fig. 1 c & d). At presentation, her right ear pain was constant, burning, throbbing and aching in nature, 9/10 on visual analogue scale (VAS) [Visual analogue scale is a reliable non-verbal measurement scale which is commonly used to assess pain and disability [11,12]. Patients are asked to draw a vertical line on a 10 cm
horizontal scale, with “0” being considered as “no pain” and “10” being “worst imaginable pain.” and was occasionally radiating to the lateral aspect of lower jaw. There were no other symptoms associated with pain. Cold breeze was the only identified aggravating factor and nothing was able to relieve her pain. Her past medical history included Meniere disease, pancreatitis, bronchitis and depression. She was taking opioids (Methadone 10 mg/tab, 6 tabs/day and oxymorphone 10 mg/tab, 1–2 tabs/3times/day), antidepressants (citalopram 20 mg/tab, 1.5 tabs/day), diazepam (5 mg/tab, 1 tab/4 times/day) and anti-emetic medications (promethazine 5 mg/tab, 1 tab/4 times/daily) at presentation. On neurological examination, higher mental functions were normal and except for absence of hearing in right ear, the rest of the cranial nerves was normal. Motor, sensory and cerebellar examination did not reveal any abnormality.

Fig. 1. (a & b) CT temporal bone and MRI with contrast images at the time of presentation to our hospital following mastoidectomy at another hospital. (c & d) CT temporal bone and MRI with contrast images at the most recent follow up following right radical mastoidectomy and obliteration of external auditory canal.

Fig. 2. X-ray lateral (a) and anterior–posterior views (b) showing the placement of an octode electrode (Medtronic, Inc., Minneapolis, MN) in the preauricular region during the trial PFNS therapy. Note the right retromastoid craniotomy defect.
Due to the refractory nature of her right ear pain and failure of all other medical and surgical treatment modalities, Neuromodulation options were considered to alleviate her pain. Following written, informed and valid consent, she underwent trial implantation of right preauricular peripheral nerve stimulator lead implantation (Fig. 2). The trial was done with percutaneous leads placed in front of the ear and attached to an external stimulator. She reported >50% pain relief in lower part of right ear. Following a successful trial of 7 days, she decided to pursue the permanent implantation with additional coverage of the jaw. Subsequently, she underwent permanent implantation of right preauricular and infra-orbital peripheral nerve stimulators and right infraclavicular pulse generator. This was done through a cervical incision with leads being placed subcutaneously in the pre-auricular and infra-orbital region. She experienced >50% pain relief with reduction in pain medications following implantation. However she was not getting adequate coverage in the upper part of her right ear and an addition preauricular lead was added and infraclavicular lead was explanted (Figs. 3 & 4). Following this implantation, she was extremely satisfied with the therapy (>50% pain relief, 4/10 on VAS). Unfortunately 3 months later, due to poor healing secondary to scars of multiple ear surgeries in this region, she had superficial infection at the site of loops of electrodes and required explantation. Following explantation, her right ear pain returned to baseline and she is looking forward to reimplantation of her system following clearance of superficial infection.

Discussion

The sensory innervation of ear is complex and involves the fifth (third division), seventh, ninth and tenth cranial nerves along with the second and third divisions of upper cervical nerve roots [4,13]. The nociceptive impulses from the inner aspect of the tympanic membrane are mediated by glossopharyngeal and vagus nerves, whereas the external portion of the tympanic membrane is mediated by trigeminal (auriculotemporal branch of mandibular division) and facial nerves [4,13]. Similarly, auditory canal is innervated by auriculotemporal and facial nerves [4,13].

Peripheral nerve field stimulation was first described by Wall and Street in 1967 [14]. In 1999, Weiner and Reed published the success of this technique in patients with occipital neuralgia which fostered the wider acceptance of this therapy [15]. Currently, PFNS has been explored for a variety of regional pain syndromes [8–10]. PFNS has been reported to cause >50% alleviation of pain on visual analogue.

Fig. 3. X-ray lateral (a) and anterior–posterior views (b) showing the placement of two octode electrodes (Medtronic, Inc., Minneapolis, MN) in the preauricular and infraorbital regions during the permanent PFNS therapy.

Fig. 4. X-ray lateral (a) and anterior–posterior views (b) showing the placement of two octode electrodes (Medtronic, Inc., Minneapolis, MN) in the preauricular region following revision PFNS therapy.
scale (VAS) in patients with refractory chronic neuropathic pain [16–20].

Indications and contraindications for PFNS

Patients with severe, chronic refractory neuropathic pain who failed all other modalities of treatment such as pharmacotherapy, trans-cutaneous nerve stimulation, trigger point injections, nerve blocks, physical therapy, Botox or acupuncture are ideal candidates for PFNS therapy [19]. The pain either should be limited in the anatomical distribution of a nerve or should be covered with combination of available electrodes. Complete absence of sensations in the area of pain is a relative contraindication for this therapy. In patients with severe allodynia, bracketing the area of pain can be useful. There should not be underlying neuropsychological issues associated with chronic pain [8,19]. Successful trial with >50% reduction in pain intensity on VAS is mandatory prior to permanent implantation. Apart from contraindications similar to other surgical procedures such as bleeding disorders, immunosuppression, active infection; those specific to PFNS include underlying psychological issues, drug seeking, inadequate family support, medico-legal issues and patients requiring frequent MRI [8,19].

Surgical techniques and programming parameters

Both trial and permanent placements of PFNS are usually performed under general anesthesia as a day care procedure. An ON-Q tunneler (I-Flow, Lake Forest, CA) is used for the placement of four or eight contact leads [Quad, Octad, Quad Plus, or Quad Compact (Medtronic, Inc., Minneapolis, MN)] in the region of previously demarcated area of pain. During the trial implantation of electrodes, the entry points are created using the Tuohy needle and ON-Q tunneler is then used to implant electrodes in the designated area of pain. The electrodes are placed in epifascial plane under fluoroscopic guidance. During permanent implantation, an incision is made (supraauricular region and cervical region as in this report) to implant the electrodes in the epifascial plane using an ON-Q tunneler and to accommodate and secure the loops of electrodes in the subcutaneous pocket at the incision site. These electrodes are then tunneled and hooked to the generator placed in a subcutaneous pocket in the infraclavicular region. Low rate (20–50 Hz), low pulse width (60–250 ms) and low amplitude (1.5–2 V) have been shown to be efficacious during PFNS programming.

Mechanisms of action

Though the leads are placed subcutaneously in front of the ear the dermalatinal stimulation is the same as the external portion of ear canal and tympanic membrane and comes from the mandibular branch of trigeminal nerve. Field stimulation in trigeminal nerve distribution is already shown to be effective in neuropathic face pain [19,20]. Guarded cathodes in preauricular leads could drive the stimulation deeper in ear. Also cross-talk between a pre and post-auricular lead can drive the stimulation coverage inside the ear. This was not possible in our patient due to extensive post-auricular scarring. Most likely explanation why peripheral stimulation provides pain relief is possibly related to gate closure at periphery and pain matrix modulation centrally [19,20].

Conclusion

Refractory deep ear pain is a complex clinical problem. PFNS can be a useful last resort modality in this cohort of patients who have exhausted all other treatment modalities. However, randomized controlled studies are required to establish the efficacy of PFNS in patients with refractory otalgia and provide hope to these difficult patients.

References