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Pain Relief With Peripheral Nerve Stimulation of Superficial Cervical Plexus: A Case Report and Literature Review

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Background: Neuropathic head and facial pain are relatively common and difficult to treat pain syndromes encountered in pain clinics throughout the world. Atypical syndromes with overlapping nerve distributions can be particularly frustrating for patients and providers. In recent years, as providers search for alternatives to medications, advances in neuromodulation technology, including spinal cord stimulation and peripheral nerve stimulation (PNS), have shown promise in treating neuropathic pain confined to specific nerve distributions, such as the greater and lesser occipital nerves. However, there is limited literature describing the use of PNS for atypical facial pain. We report the first known case of PNS to the superficial cervical plexus in a 45-year-old woman with bilateral ear and postauricular pain.

Case Report: A 45-year-old woman presented with over 6 years of bilateral ear and periauricular pain in the auriculotemporal and lesser occipital nerve distribution. Conservative management, including neuropathic pain agents, ketamine infusions, nasal sprays, opioids, nonsteroidal anti-inflammatory medications, ice, heat, and transcutaneous neurostimulation, provided limited relief. She had excellent pain control with a series of nerve blocks to the superficial cervical plexus; however, relief was short term. In an attempt for a more long-term solution, she underwent implantation of the SPRINT PNS (MicroLead, SPR Therapeutics, Cleveland, OH), which resulted in 100% pain relief, improved function at work, and improved sleep tolerance throughout the 60-day treatment. Unfortunately, her symptoms returned shortly after treatment course completion.

Conclusions: To our knowledge, this is the first case utilizing PNS of the superficial cervical plexus to treat atypical facial pain. The 60-day trial resulted in 100% pain relief of previously debilitating and functionally limiting pain symptoms during the treatment phase suggesting that the superficial cervical plexus may be a reasonable target for permanent PNS implants.

Key words: Peripheral nerve stimulator, peripheral nerve stimulation, superficial cervical plexus, atypical facial pain, anesthetic blockade, pain relief, neuropathic pain, neuromodulation, SPRINT

BACKGROUND

Anesthetic blockade of the superficial cervical plexus is often utilized to provide anesthesia for surgeries of the head and the neck. We describe a technique utilizing PNS targeting this plexus for pain relief. This case report did not require Institutional Review Board approval.

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CASE PRESENTATION

A 45-year-old woman with a history of mitochondrial disease, neck pain, posttraumatic stress disorder, and hypothyroidism was referred by neurology with > 6 years of bilateral ear and periauricular pain in the auriculotemporal and lesser occipital nerve distribution (Fig. 1) (1). Her pain was described as intermittent, throbbing, aching, burning, and stabbing in nature, and exacerbated by contact when sleeping on her side or wearing work-required headsets/headphones (Visual Analog Scale 10). Her symptoms were intractable to conservative management, including first-line neuropathic pain agents, ketamine infusions and nasal sprays, opioids, and nonsteroidal anti-inflammatory medications. Adjuvant modalities, including ice and heat, were ineffective, while transcutaneous electrical nerve stimulation provided some relief for her pain symptoms.

The patient underwent a series of nerve blocks to the superficial cervical plexus with excellent, but short duration pain relief of about 2 weeks. She subsequently desired a long-term solution for her pain symptoms and underwent placement of a percutaneous peripheral nerve stimulation (PNS) device (SPRINT MicroLead, SPR Therapeutics, Cleveland, OH), which resulted in 100% pain relief, improved function at work, and decreased hyperpathia with sleeping and wearing headsets throughout the 60-day treatment that resulted in persistent symptom relief beyond removal of stimulator leads.

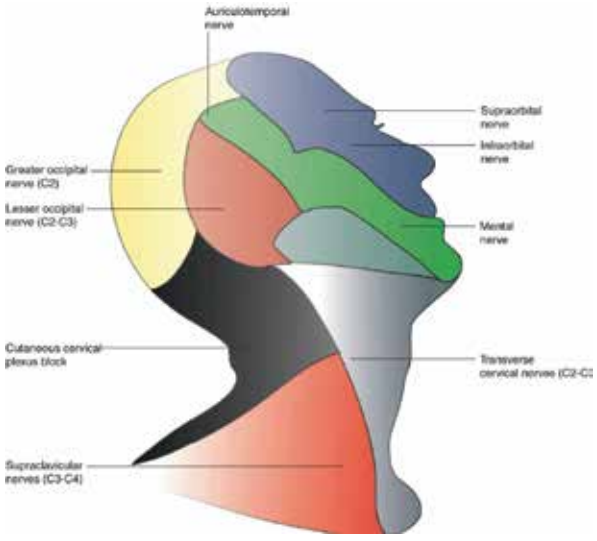

Fig. 1. Dermatomes of nerves innervating lateral face, head, and neck.

Anatomy

The superficial cervical plexus originates from the C2-C4 nerve roots, which combine to form 4 terminal branches (i.e., lesser occipital, greater auricular, transverse cervical, and supraclavicular nerves). The plexus emerges just posterior to the sternocleidomastoid muscle near the intersection of the muscle with the external jugular vein and superficial to the prevertebral fascia. The nerves go on to provide cutaneous sensation to the anterolateral neck and the anteauricular and retroauricular areas, as well as the skin overlying and immediately inferior to the clavicle on the chest wall (Fig. 2) (1).

Procedure

Informed consent was obtained from the patient prior to the start of the procedure. The patient was placed in the right lateral decubitus position and padded to foster comfort. Appropriate skin and bony landmarks were identified and pertinent vascular structures were located. The skin overlying the left lateral neck was prepped and draped in sterile fashion (Fig. 3). Ultrasound was used to identify appropriate landmarks and the location of the superficial cervical plexus (Fig. 1) (1). The skin around the planned entry point and the subcutaneous tissues were injected with 2% lidocaine local anesthetic.

A percutaneous sleeve and stimulating probe lead introduction system were assembled, inserted, and advanced along the intended course avoiding vascular structures to the superficial cervical plexus, taking care to maintain the proper depth of insertion as the introducer was advanced under ultrasound guidance. The introducer needle was delivered to a location in proximity to the nerve.

Multiple stimulation parameters were used to deliver stimulation to the left superficial cervical plexus in concert with stimulation at multiple positions around the nerve. Nerve target acquisition was confirmed by noting generation of paresthesias in the targeted nerve distributions. Various electrical parameter combinations were tested, and the lead location was adjusted (i.e., physically relocated) until the patient indicated paresthesia overlapping the distribution of the patient's typical region of pain.

The stimulating probe was removed from the introducer and a percutaneous lead was guided through the needle and delivered to a location in similar proximity to the nerve. Final location was verified

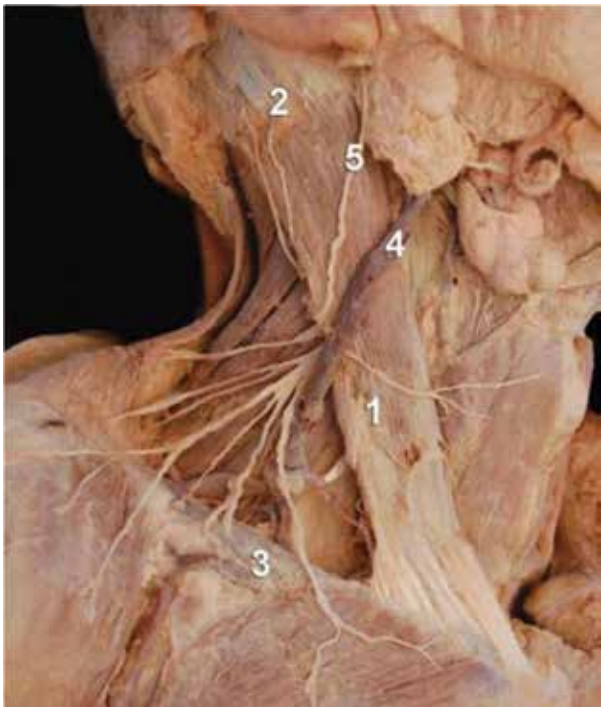

Fig. 2. The superficial cervical plexus emerges just posterior to the sternocleidomastoid muscle (#1) near the intersection of the muscle with the external jugular vein (#4) and superficial to the prevertebral fascia. The nerves provide cutaneous sensation to the anterolateral, anteauricular, and retroauricular areas of the neck.

with electrical stimulation and documented with ultrasound (Fig. 4).

The introducer needle was removed, and the exposed end of the percutaneous lead was attached to an external stimulator unit. Various electrical parameter combinations were again tested. After confirming that the lead impedance was in the normal range, the external stimulator unit was detached, the needle was removed, and the lead was anchored at the skin.

The lead was threaded into the connector block and electrical continuity and desired patient response was confirmed. The connector block was attached to the external stimulator unit.

The site was covered with a sterile occlusive dressing and an ultrasound image was taken to document the final placement (Fig. 4). The patient was observed for stability of vital signs and comfort.

Device

SPRINT MicroLead with OnePass Introducer.

Fig. 3. Preprocedure setup.

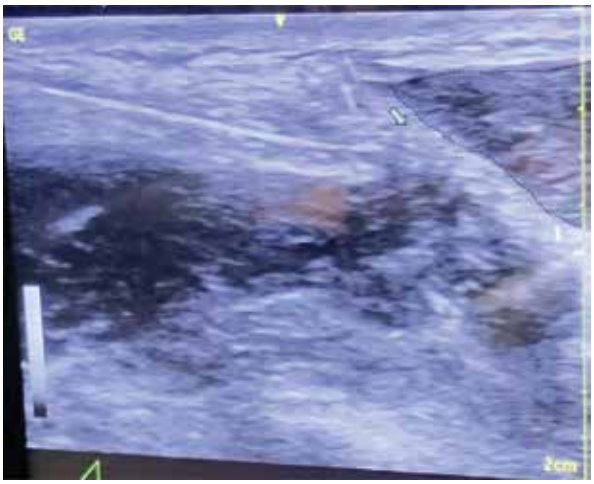

Fig. 4. Ultrasound figure with arrow demonstrating needle advancement to superficial cervical plexus. Stimulation provided pain relief, confirming the target area and appropriate coverage of pain distribution.

DISCUSSION

In patients with mitochondrial disease, chronic pain is fairly common and most often is neuropathic in nature (2). In our patient, she had pain throughout her body that was likely neuropathic based on history and physical exam; however, her neck and periauricular pain was the most debilitating area as it impacted her sleep and work life. The distribution of her pain was overlapping nerves stemming from the superficial cervical plexus; therefore, we targeted this area.

To our knowledge, this is the first case of PNS ap-

plied to the superficial cervical plexus. The 60-day trial resulted in 100% pain relief of previously debilitating and functionally limiting pain symptoms with pain relief continuing post-lead removal pain control at > 50%.

The use of PNS has become more common in the treatment of many neuropathic pain conditions. The mechanism by which PNS provides pain relief is not completely understood; however, many studies have attempted to identify a mechanism of action. The initial explanation for the mechanism is based on the gate control theory, initially proposed by Melzack et al (3). Many studies (4,7) since have identified multiple areas that are affected by stimulation of the nervous system, including modulation of peripheral, central, autonomic, and inflammatory pathways. Peripherally, PNS is thought to disrupt the transmission of nociceptive afferent Aδ and C fibers (4,9). Centrally, changes in neuropeptides and neurotransmitters, including serotonergic, dopaminergic, GABAergic, and glycinergic pathways (5,7), as well as changes in concentrations of substance P and calcitonin gene-related peptide (6) have been identified. One study (8) has shown supraspinal changes, including activation of the dorsal lateral prefrontal cortex and descending inhibitory pain pathways, including the anterior cingulate cortex and parahippocampal area.

Robust nonnociceptive input directly to the focal areas of pain where the PNS is selectively targeted reduces the severity of pain perceived by reconditioning the central nervous system from the periphery. This remote targeting is theorized to allow more selective activation of large diameter afferent fibers (9). This type of selective reconditioning cannot be obtained by nerve blocks or radiofrequency ablations as there is a passive deprivation of nociceptive input. When there is robust focal activation of the peripheral target nerves, there is potential for cortical input to induce activity-dependent remapping and sustained analgesia, even long after removal of the PNS (9).

CONCLUSIONS

Neuropathic pain is often difficult to treat and many patients fail first-line conservative therapy and medications that have noticeable side effects; therefore, limit their utilization. We present the first documented case using a PNS device to target the superficial cervical plexus providing 100% relief of previously debilitating postauricular pain after the 60-day trial and > 50% pain relief post-lead removal. This case report demonstrates additional use of minimally invasive peripheral neuromodulation in the treatment of neuropathic pain, suggesting that the superficial cervical plexus may be a reasonable target for PNS implants.

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