

Case Report

Successful Treatment of Refractory Pudendal Neuralgia with Pulsed Radiofrequency

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Pudendal neuralgia (PN) involves severe, sharp pain along the course of the pudendal nerve, often aggravated with sitting. Current therapies include medication management, nerve blocks, decompression surgery, and neuromodulation. The ideal management for PN has not been determined.

We present a case of a female with 1.5 years of sharp, burning pain of the left gluteal and perineal regions. She could not sit for longer than 10 to 15 minutes. Sacroiliac joint, epidural, and piriformis injections did not improve her pain. She had tried physical therapy, occupational therapy, massage, and acupuncture but the pain persisted. Medication treatment with oxycodone-acetaminophen, extended release morphine sulfate, amitriptyline, and gabapentin provided only minor relief and she had failed other multianalgesic therapy. She had been unable to work at her desk job for over a year. She had a positive response to 2 diagnostic pudendal nerve blocks with lidocaine that provided pain relief for several hours. This patient elected to undergo pulsed radiofrequency (PRF) of the left pudendal nerve in hopes of achieving a longer duration and improved pain relief. PRF was carried out at a frequency of 2 Hz and a pulse width of 20 milliseconds for a duration of 120 seconds at 42 degrees Celsius. After the procedure she reported tolerating sitting for 4 to 5 hours. Her multianalgesic therapy was successfully weaned. At 5 months follow-up she felt motivated to return to work. One and a half years after the procedure the patient is only taking oxycodone-acetaminophen for pain relief and still has good sitting tolerance. There were no procedure-related complications.

To our knowledge PRF for the treatment of PN has not been reported elsewhere in the literature. PRF is a relatively new procedure and is felt to be safer than continuous radiofrequency. Current literature suggests that PRF delivers an electromagnetic field, which modifies neuro-cellular function with minimal cellular destruction. We conclude that PRF of the pudendal nerve offers promise as a potential treatment of PN that is refractory to conservative therapy.

Key words: pudendal neuralgia, pulsed radiofrequency, pudendal nerve, neuromodulation

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Pudendal neuralgia involves severe sharp pain along the course of the pudendal nerve, which is often aggravated with sitting (1,2). This condition is also characterized by sensations of burning, heaviness, and presence of foreign objects in the vagina

or rectum (2). Onset may occur postoperatively, after repetitive bicycling, or without a provoking factor. Current therapies include medication management, pudendal nerve blocks, decompressive surgery, and neuromodulation with implanted pulse generators

(1). The ideal management of pudendal neuralgia has not yet been defined (3).

Pudendal neuralgia significantly impacts a patient's quality of life (1). These patients struggle with activities of daily living which involve sitting, such as working at desk jobs or riding in a car. This type of chronic pain reflects sensitization (wind-up) and altered processing of pain signals by the peripheral and central nervous systems that can lead to physical and psychosocial impairments as well as secondary pain-related behaviors that can lead to decreased functional status and feelings of self-worth.

The pudendal nerve is comprised of the terminal branches of the sacral plexus: S2, S3, and S4 (2,4). This is a mixed nerve, that provides sensory innervation of the lower parts of the vagina, vulva, and perineum as well as motor innervation of the perineal muscles (4). The course of the pudendal nerve predisposes it to impingement at 3 common locations: between the sacrotuberal and sacrospinous ligaments, within the pudendal canal, and crossing the faciform process of the sacrotuberal ligament (2).

We present a case of a female with 1.5 years of sharp, burning pain of the left gluteal and perineal region. She could not sit for longer than 10 to 15 minutes due to neuralgic pain. This patient had failed conservative therapy with muscle strengthening exercises and use of a doughnut. She had a positive short-term response to 2 diagnostic left pudendal nerve blocks. This patient elected to undergo pulsed-wave radiofrequency (PRF) of the left pudendal nerve in hopes of an improved and longer duration of pain relief.

Continuous radiofrequency ablation (CRF) is a percutaneous minimally invasive technique that has been in clinical use for over 25 years (5). PRF is a more recent neuromodulatory technique that is felt to be safer than conventional CRF (6). Although the exact mechanism of action for PRF is unknown, current literature supports involvement of electromagnetic fields resulting in neuromodulation. Our clinical experience as well as recent literature suggests that PRF may be useful in treatment of refractory neuropathic painful conditions (2,6-16). We have successfully treated several cases of lateral femoral cutaneous neuralgia and ilioinguinal neuralgia with PRF, which gave us the impetus to try PRF of the pudendal nerve, which has not been reported elsewhere in the literature.

CASE REPORT

A 41-year-old woman was referred to our office with sharp, burning pain for approximately 1.5 years of the left gluteal and perianal region. Her past medical history was significant for migraines. The onset of pain occurred while she was climbing stairs with heavy luggage. Sitting or lying on the gluteal area exacerbated the pain. Pain was also intensified by full meals, bowel movements, and contact with clothing. She reported difficulty passing stools and feeling bloated. She was only able to sit for a maximum of 10 to 15 minutes. She rated the pain at 9/10 at maximum intensity. Sitting on the toilet, standing, and walking were alleviating factors. She was unable to work at her desk job for more than a year secondary to her inability to sit.

Because pudendal neuralgia is frequently misdiagnosed, she had previously been treated with sacroiliac joint, epidural steroid, and piriformis injections that did not improve her pain. She had failed multianalgesic therapy with extended release morphine sulfate, hydrocodone-acetaminophen, oxycodone-acetaminophen, nabumetone, tizanidine, amitriptyline, and gabapentin. Despite physical therapy, occupational therapy, massage, TENS unit, and acupuncture, her pain persisted. She had also visited a chiropractor and a neurologist.

Two transvaginal diagnostic nerve blocks of the left pudendal nerve were performed. The patient was positioned in the dorsal lithotomy position. The ischial spine and attachment of the sacrospinous ligament were identified by transvaginal palpation. At 1.5 cm medial to the left ischial spine 15 mL of 1% lidocaine was injected using a 5-inch needle from the BD pudendal nerve block kit. Pain was rated as 9/10 prior to the procedure and 2/10 post procedure. She experienced significant pain relief for 3 hours. During this 3-hour interval she was able to sit without pain. The second diagnostic left pudendal nerve block was performed 2 weeks later. The same technique was used with 8 mL of 2% lidocaine injected. Pain was described as 8/10 prior to procedure and 4/10 post procedure. Pain relief again lasted several hours.

Given the duration of her symptoms, it was expected that the patient experienced peripheral pudendal neuralgia and central pain due to spinal cord sensitization (wind-up). However, given that the patient had 2 successful diagnostic left pudendal nerve blocks, it was believed that the patient's pain presentation was predominantly of peripheral etiology and

any further residual central sensitization component could be lessened by treating the underlying peripheral lesion by PRF. Therefore, PRF of the left pudendal nerve was proposed as a treatment option and the patient decided to proceed in hopes of achieving a longer duration and improved pain control.

The patient was positioned supine with her knees flexed and the plantar aspects of her feet conjoined. She was prepped and draped in a sterile fashion. The ischial spine and attachment of the sacrospinous ligament were identified transvaginally. At 1.5 cm medial to the left ischial spine a 22-gauge 4 mm active tip RF needle was advanced approximately 2 cm incrementally near the left pudendal nerve. The impedance was recorded at 350 ohms and 0.25mV of sensory stimulation performed at 50 Hz produced paresthesias in the distribution of the pudendal nerve. PRF lesioning of 2 Hz with 20 millisecond pulses was performed at 42 degrees Celsius for 120 seconds. The patient tolerated the procedure well without any complications.

This patient was being treated with gabapentin, amitriptyline, oxycodone-acetaminophen, and extended release morphine sulfate at the time of PRF. Post procedurally, pain improved significantly and her function progressed. She was now able to tolerate prolonged sitting for 4 to 5 hours. Morphine sulfate extended release, amitriptyline, and gabapentin were discontinued. Five months after the procedure, the patient felt motivated to return to work. At 6 months, post procedure patient reported significant improvement in her pain and good sitting tolerance. At 1.5 years post procedure, the patient only takes 3 tablets of oxycodone-acetaminophen per day. She is able to tolerate 4 to 5 hours of sitting per day.

Discussion

The ideal management of pudendal neuralgia has not been defined (3). A recent retrospective study concluded that pudendal neuralgia is "poorly recognized and poorly treated" (1). Results showed that most patients had slight to moderate improvement in pain with conservative therapy. Only 31% improved after pudendal nerve blocks and 60% improved after surgical decompression. Two patients improved with peripheral neuromodulation of the pudendal nerve with a pulse generator. The authors suggest that neuromodulation be further investigated because current therapeutic options for patients with pudendal neuralgia are "less than desirable" (1).

The pudendal nerve is typically blocked by iden-

tifying the ischial spine which can be palpated either transvaginally or per rectum. It is important to use a needle with a guide in order to limit the depth of submucosal penetration. When a left sided block is performed, the ischial spine is palpated with the index finger of the left hand, the syringe is held in the right hand, and the needle is guided between the index and middle fingers of the left hand towards the ischial spine. The sacrospinous ligament lies 1 cm medial and posterior to the spine. The needle is passed through the ligament for a distance of 1 cm until a loss of resistance is appreciated. At this point the tip lies in the area of the pudendal nerve. The pudendal vessels are closely associated. After aspiration, the local anesthetic solution is injected.

In 1998 a review of RF suggested that it may be effective for the relief of perineal pain. However the authors stress that low temperature PRF lesions are preferred to avoid potential sexual, bowel, or bladder dysfunction as well as neuritis and/or deafferentation syndrome that may occur with CRF (6).

The RF lesioning technique involves placement of an insulated needle with an active tip in the vicinity of a nerve or ganglion (5). A grounded electrode is passed through the cannula and RF current is emitted at the tip of the needle (9).

There are 2 types of RF lesioning that are used clinically: CRF and PRF. CRF is the conventional method which uses a constant output of high frequency current and produces temperatures > 45 degrees Celsius (5). The heat production associated with this technique is neuroablative. Alternatively, PRF uses brief pulses of high voltage electric current (5). Pauses between the pulses allow heat to dissipate and thus less nerve destruction occurs (5). The temperature with PRF generally does not exceed 42 degrees Celsius (5).

PRF was first developed in 1995 and its first clinical application took place in February 1, 1996 (17). The exact mechanism of action of PRF is unknown (18). Initially the proposed mechanism of action of pain relief was that heat produced by RF caused destruction of the nerve. However, the observation that sensory loss was transient while pain relief was of much longer duration lead to the hypothesis that temperature was not the only mechanism of action responsible for changes in pain perception (5). It has been proposed that PRF may act by modulating pain perception rather than directly destroying neural tissue (16). Although PRF was originally described as nondestructive, this has been challenged. It may be premature to describe

PRF as nondestructive when destructive effects would be expected to occur at a microscopic level that have yet to be well examined (19). The most likely causes of RF pathologic lesions are heat, high electric fields, and high current fields. Heat is associated with a rapid spread of energy among all tissues, even at a molecular level. Electric fields induce forces on charged molecular structures that may result in their distortion and dislocation as well as disruption of cellular function. For example electric fields may be associated with ion conduction across the neuronal membrane, which may alter resting and threshold potentials of neurons and ultimately mediate the perception of pain. Electroporation is a process induced by transmembrane potentials that creates pores and possibly leads to rupture of the cell membrane. Thus electroporation may cause increases in membrane permeability associated with cell stress and death. Electroporation has been proposed as a possible mechanism of action for PRF. High current fields may also affect cellular structure by causing collisions of ions and molecules. Tissue damage at a microscopic level after PRF needs to be further evaluated.

One study looked at exposure of the cervical dorsal root ganglion (DRG) to both CRF and PRF (20). C-Fos expression was found to increase in the dorsal horns of rats bilaterally 7 days after intervention with both CRF and PRF. C-Fos is a nonspecific immediate early gene marker that is used to detect activated neurons. This supports that c-Fos activation is heat independent and may provide information as to the mechanism of action of PRF in pain relief. Though the exact mechanism of action of PRF is unknown, current literature suggests that electromagnetic fields lead to the neuro-modulation as evidenced by altered c-Fos expression.

Another recent study challenges the common perception that PRF is a "nonthermal" and "nondestructive" lesion (19). Field predictions of both CRF as well as PRF were made. The authors determined that CRF produced heat that destroyed neurons and described PRF as producing heat in bursts that is in the range associated with neurodestruction. PRF was associated with "very high electric fields that may be capable of disrupting neural membranes and function" and some of this neuroablation may be temperature dependent. A small area of tissue destruction after PRF that may be attributed to heat spikes is described by these authors (18,19). It is uncertain whether this destruction is responsible for the clinical

effects of PRF.

Another study looked at cell stress after PRF by studying activating transcription factor 3, a marker of cellular stress, in rats (21). Results showed that PRF appears to be selective as it targets neurons whose axons are small in diameter, specifically C and A-delta nociceptive fibers. This is consistent with the absence of sensory and motor deficits following PRF. The authors feel it is "reasonable to assume that cell stress will result in general reduction of cellular activity including down-regulation of excitatory neuromodulators within nociceptors and this may underlie behavioral analgesia" (21).

To date only one randomized controlled trial has been published on PRF wherein PRF of the cervical DRG was performed on patients with chronic cervical radicular pain (15). At 3 months, PRF showed significantly better outcomes than placebo and at 6 months those treated with PRF had a significant decrease in need for pain medication. No complications related to PRF were reported. However, because of low inclusion rates these results are difficult to apply to clinical practice. The authors concluded that PRF of the cervical DRG might provide pain relief for carefully selected patients with chronic cervical radicular pain.

Numerous case reports and case series of PRF of numerous peripheral nerves and ganglions for treatment of chronic pain can be found in the literature (2,6-16). Beneficial clinical results are found after PRF of the obturator nerve, femoral nerve, medial and lateral branches of the dorsal horn, stellate ganglion, supraclavicular nerve, DRG, S1 nerve root, the gasserian ganglion, glossopharyngeal nerve, sphenopalatine ganglion, ilioinguinal nerve, iliohypogastric nerve, genitofemoral nerve, supraorbital nerve, frontal nerve, and lateral femoral cutaneous nerve. No adverse effects of PRF were reported in any of these cases. Effects of pain relief appear to be variable ranging from 2 to > 30 months with a mean duration of 9.2 months in one case series of PRF of the cervical DRG (13).

An additional benefit of PRF is that the procedure can be repeated if pain recurs because minimal tissue has been destroyed (14,21). PRF is minimally invasive, well tolerated, and lacks potential adverse effects associated with high temperatures (8) and thus holds promise in patients with chronic neuralgic pain that is refractory to conservative therapies.

Lack of randomized controlled trials and known mechanism of action are recognized limitations. A

2006 review of the literature concluded that evidence is accumulating which suggests efficacy and that PRF does have a place in clinical practice (22). It is noted that CRF and PRF should be considered different and nonequivalent procedures (23). PRF requires less precision, less time, and is associated with much lower risk than CRF. PRF is proposed with the potential of being a safer method of neuromodulation and possibly expanding the indications for clinical use with equal results to CRF (18).

One potential downfall of the diagnostic nerve block and the described technique is that the pudendal nerve is a difficult block to perform and master. This difficulty was described in obstetric patients treated with bilateral pudendal blocks during labor (24). In only 36 of 100 patients was positive bilateral pudendal nerve blockade achieved. This is important because unless patients have a positive response to diagnostic nerve blockade they would not be candi-

dates for PRF and a failed nerve block may be related to the experience or skill of the practitioner.

PRF of the pudendal nerve may potentially offer promise as a treatment for pudendal neuralgia that has failed conservative therapy. Our patient had pain control for an increased duration than previously gained from nerve blockade. Further investigations are needed to determine the exact mechanism of action of PRF.

CONCLUSION

PRF of the pudendal nerve may be safe and beneficial for a select group of patients who have obtained consistent relief with pudendal nerve blocks. PRF lesioning of the pudendal nerve may safely provide relief in patients suffering from intractable perineal pain.

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