

Case Report



Sacral Neuromodulation as a Treatment for Pudendal Neuralgia

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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: 04-15-2014
Revised manuscript received: 05-14-2014
Accepted for publication:
06-09-2014

Free full manuscript:
www.painphysicianjournal.com

Pudendal neuralgia is a debilitating pain syndrome, and finding long-lasting treatment modalities has been challenging in pain management. The pudendal nerve has sensory and motor functions, and influences autonomic functions. Thus, entrapment or damage of this nerve can have multiple serious implications. The constellation of symptoms which result from injury to this nerve is commonly referred to as pudendal neuralgia. When conservative therapy does not provide adequate pain relief and surgical procedures fail or are not viable options, central and peripheral nerve stimulation can be effective treatment modalities. More recent approaches to treatment include the use of peripheral nerve stimulation through the use of an electrical lead placed next to the pudendal nerve in the ischioanal fossa. Also, epidural stimulation of the conus medullaris and pulsed radiofrequency ablation of the pudendal nerve have been shown to be effective in small patient populations. We present the case of a 36-year-old woman who sustained pudendal nerve injury during a hysterectomy and subsequently developed intractable pelvic pain and pudendal neuralgia. Conservative treatment measures failed, but she obtained excellent results from peripheral nerve stimulator therapy. Permanent implantation consisted of 4 tined Interstim leads, individually placed into the bilateral S3 and S4 foramina. The patient has been followed for approximately 4 years since her procedure, demonstrating increased function as she is able to stand and sit for prolonged periods of time. She has returned to her usual daily activities, including horseback riding. This is the first reported case of transforminal sacral neurostimulation providing excellent relief of pudendal neuralgia related symptoms.

Key words: Pudendal neuralgia, sacral neurostimulation, peripheral nerve stimulator, pelvic pain, nerve stimulation, interventional pain procedures, surgical management of pain

Pain Physician 2014; 17:E645-E650

The pudendal nerve is derived from sacral S2-4 nerve roots, has mixed sensory and motor functions, and influences the sympathetic nervous system subserving pelvic organs. It has 3 terminal branches: the inferior rectal nerve, the perineal nerve, and the dorsal nerve that provides innervation

to the clitoris or penis. It supplies motor control of the external anal and external urethral sphincters and the pelvic floor muscles; provides sensory innervation to the anal, perineal, and genital regions; and influences the sympathetic nerve supply to the pelvis. As such, entrapment or compression of this nerve can cause

bowel, bladder, and sexual dysfunction, in addition to pain in the aforementioned regions. This clinical picture is termed pudendal neuralgia, and has been documented in medical literature dating back to its initial description by Boisson et al in 1966 (1). Though there are diagnostic modalities that can be suggestive of pudendal neuralgia, it is a clinical diagnosis with specific criteria.

There is no clear consensus on the best treatment of this condition (2). Conservative approaches include physical therapy, anticonvulsants (including gabapentin or pregabalin), opioids, muscle relaxants, and tricyclic antidepressants, but no randomized controlled trials have been done to investigate the efficacy of these medications (3,4). Pudendal nerve injections have been shown to have an efficacy of 31% – 62% (5). Surgical decompression of the pudendal nerve has also been attempted, with success rates as high as 60% – 71% (5,6), but other studies suggest lower rates of treatment efficacy (6).

More recent approaches to treatment include the use of peripheral nerve stimulation through the use of an electrical lead placed next to the pudendal nerve in the ischioanal fossa. This approach was 100% successful in a study of 2 patients (5). Epidural stimulation of the conus medullaris and pulsed radiofrequency ablation of the pudendal nerve have also been shown to be effective in small patient populations (2).

The preliminary success of these neuromodulatory techniques is encouraging and suggests that neuromodulation may be an effective technique to provide long-term pain relief to patients suffering from pudendal neuralgia. In this report, we will present the case of a woman with pudendal neuralgia who, after failing conservative treatments, has been able to achieve prolonged, significant improvement of her symptoms following the percutaneous insertion of permanent nerve stimulation leads through the sacral foramina. This is a novel and first reported use of this technique for the treatment of pudendal neuralgia and represents another potential treatment modality for this difficult-to-treat condition.

CASE REPORT

The patient is a 36-year-old, gravida 1 para 1, woman who underwent robotic-assisted radical hysterectomy for a Stage 1B adenocarcinoma of the cervix approximately 12 weeks prior to presentation at the Pain Management Center. She was experiencing severe pelvic pain attributed to intraoperative injury of the

right pudendal nerve. A burning, sharp pain was rated as 10 out of 10 on the Numerical Rating Scale (NRS). It would decrease to 0 out of 10 when supine, but sitting or standing even for short periods of time would cause the pain to again increase to 10 out of 10. While most of the pain involved the right sides of the vaginal wall, rectum, labia, and clitoris, she also had deep, achy midline pain just anterior to the sacrum and involving the vaginal wall at that location. At presentation to the pain clinic she was on 80 mg of Oxycontin TID, pregabalin 150 mg TID, baclofen 10 mg TID, desipramine 50 mg at night, and ibuprofen 600 mg every 6 hours as needed. Despite these medications, her pain limited her function dramatically, and, as a result of her inactivity, she had gained over 10 kilograms in the span of 2 months.

Of note, the patient had a magnetic resonance neurography that demonstrated apparent tension on the right S3 root proximal to the pudendal nerve resulting in pudendal neuralgia. She subsequently had multiple interventions including caudal epidurals, S3/S4 nerve root blocks, a trial of a continuous transforminal local anesthetic infusion of the right S3 root, and pulsed radiofrequency neuromodulation of S3/S4. These interventions provided transient benefits, save for the local anesthetic infusion, which provided sustained relief of the right-sided symptoms. Midline symptoms that crossed over to the left side persisted, and the patient found the numbness associated with the infusion bothersome. At this juncture, the patient was willing to explore other therapeutic options that would decrease her pain, but, more importantly, allow her to regain her function in order to participate in her daily activities.

Given the efficacy of the continuous S3 root local anesthetic infusion, we offered the patient a trial of bilateral pudendal nerve stimulation. The risks, benefits, and alternatives of sacral nerve stimulation (SNS) were discussed at length with the patient. On the day of the stimulator trial, she was placed in prone position, prepped, and draped as appropriate for bilateral S3 foramina nerve stimulator insertion. Omnipaque dye was used to confirm root location, and 2 4-electrode leads were advanced under direct fluoroscopic guidance to appropriate anatomical points bilaterally. Intraoperative testing caused the expected paresthesias in the distribution where pain was most intense. During the 5-day trial, she received excellent pain relief, including while standing and walking. Whereas before she was unable to stand for more the 15 minutes, she walked

multiple blocks around downtown with the stimulator in place, which also significantly improved her mood. She reported greater than 75% relief during the trial period. Unfortunately, she had residual pain in her perineum, which was likely suggestive of S4 involvement.

One month after this, the patient was brought to the operating room for permanent nerve stimulator implantation. After anesthetic induction, the patient was placed prone, prepped, and draped in appropriate sterile fashion. Needles provided by Medtronic were introduced using a transforaminal approach into the S3 and S4 foraminal openings bilaterally under fluoroscopic guidance. A total of 5 mL of Omnipaque dye were injected and showed good delineation of the bilateral S3 and S4 pudendal nerve roots (Figs. 1 and 2). Once needle positioning was found to be satisfactory, stimulator leads were placed into the foraminal openings and advanced transforaminally (Fig. 3). The correct positioning of the 4 leads was confirmed by anteroposterior and lateral fluoroscopy. There were 4 electrodes per lead. The patient was awoken, and stimulation at each foraminal opening was conducted. The patient corroborated that there was appropriate paresthesia along the nerve distribution corresponding to the nerve being stimulated by each electrode. After finding the lead positioning to be satisfactory, the patient was anesthetized again. A 15-blade scalpel was used to make small incisions at each of the needles to facilitate tunneling of the 4 leads to the generator, which was positioned over the right ilium. Extensions were used to reach the neurostimulator, and leads were carefully protected and secured with 2.0 silk suture. The satisfactory positioning of the electrodes after tunneling was confirmed using AP and lateral view on fluoroscopy. In summary, permanent implantation consisted of 4 tined Interstim (Medtronic, Minneapolis, MN, #3889)



Fig. 1. Positioning of spinal needles in the bilateral S3 and S4 sacral foraminal openings.



Fig. 2. Lateral view of the needle positioning in the foraminal openings. Clear delineation of the S3 and S4 nerve roots can be seen.



Fig. 3. Positioning of nerve stimulator leads in the bilateral S3 and S4 sacral foraminal openings.

leads, individually placed into the bilateral S3 and S4 foramina. Two 20 cm bifurcated extensions (Medtronic, Minneapolis, MN, #37082) were tunneled to where the Restore Ultra pulse generator (Medtronic, Minneapolis, MN, #37712) was secured. With the additional S4 leads in place, the patient claimed concordant pain relief.

The patient has been followed for almost 4 years since her procedure, with excellent increase in her function as she is able to stand and sit for prolonged periods of time. She has returned to her usual daily activities, including horseback riding. She requires only 40 mg of Oxycontin twice daily, and she has been able to decrease doses of other pain medications. During subsequent visits to the pain management center, she rated her pain as 2/10 on the NRS. The patient's mood has also dramatically improved. Moreover, she and her husband are extremely grateful that she is able to function and resume most of her normal activities of daily living.

Discussion

Pudendal neuralgia is a neuropathic condition involving the pudendal nerve that causes severe, sharp pain in the genital area (4). The pudendal nerve is prone to entrapment or compression between the sacrospinous and sacrotuberous ligaments at the ischial spine and within the pudendal (Alcock's) canal (7). Entrapment of the nerve can cause severe neuropathic pain and is often accompanied by bowel, bladder, and sexual dysfunction. The most common causes of pudendal nerve entrapment in women are childbirth, pelvic trauma, and surgical injury. Pelvic trauma is the most common cause in men (4). Pudendal neuralgia is estimated to affect 1% of the general population, and it is more common in women than men. Because it is difficult to diagnose, neurophysiologic findings can be normal, and practitioners often fail to consider it in their differential diagnosis, the mean time to diagnosis is 4 years, and the average number of physicians consulted before diagnosis ranges from 10 to 30 (8).

Labat et al (9) published the Nantes Criteria in 2008 to help to make the diagnosis of pudendal neuralgia. The criteria include:

1. Pain in the anatomical territory of the pudendal nerve
2. Pain is worsened by sitting (though a distinguishing characteristic of this syndrome is that the pain is improved when sitting on the toilet or a donut pillow)
3. The patient is not woken at night by the pain

4. No objective sensory loss on clinical examination
5. Pain relief with anesthetic pudendal nerve block.

Exclusion criteria for neuropathy included purely coccygeal, gluteal, or hypogastric pain, exclusively paroxysmal pain, isolated pruritus, and the presence of imaging abnormalities able to explain the symptoms. Pudendal neuralgia is a clinical diagnosis.

There is no universally agreed upon approach to treatment of pudendal neuralgia (2). Treatment options have historically been limited and had variable results. Even in cases of pudendal neuralgia due to identified nerve entrapment where surgical decompression is an option, the European Association of Urology in its 2012 Guidelines on Pelvic Pain found that pain relief from surgery only occurs 66% of the time if pain has lasted less than 6 years, and 40% of the time if pain has existed for greater than 6 years (10). A study in 2007 that followed patients for 4 years after transperineal, transgluteal, or transchiorectal decompressive surgery of the pudendal nerve found that only 50% of patients had some relief, but no comparison could be made to control patients because they had not received follow-up (6). Pharmacologic therapies include nonsteroidal anti-inflammatory drugs (NSAIDs), tricyclic antidepressants, anticonvulsants, muscle relaxants, and opioids (3). A retrospective study in 2005 showed that conservative treatment with medications and sitting pads provided slight to moderate improvements in pain (5). To achieve the same degree of slight to moderate relief, pudendal nerve blocks were successful in only 31% of patients while surgical decompression was effective in only 60% of patients (5). Physical therapy targeting pelvic floor musculature has also been employed, but the efficacy of physical therapy for this syndrome is unclear (4).

Spinal cord stimulation of the conus medullaris was shown to be effective in a prospective trial where 20 of 27 patients were positive responders, and, of those 20, 100% had long-term relief (11). Though spinal cord stimulation has proven helpful in chronic pelvic pain, the optimal location for spinal cord stimulator placement is unclear. Placing the stimulator at the conus would appear most logical, but anatomical structures and the movement of the conus may contribute to the occasional failure of this treatment modality (12). We describe an intervention that bypasses this problem by directly stimulating the involved nerves peripherally. Another study showed peripheral neuromodulation through the use of a pulse generator attached to a

tined quadripolar lead next to the pudendal nerve at the ischioanal fossa provided 100% relief, but the study only had 2 patients (5). A case report in 2009 of 3 patients using a similar technique (except with additional neurophysiology guidance during lead placement) also showed that peripheral pudendal nerve neuromodulation can be effective (3). A 1998 review of radiofrequency ablation (13) suggested that radiofrequency may be helpful in treating perineal pain, and a recent case report in a single patient with pudendal neuralgia demonstrated significant improvement in pain and functional status following treatment with pulsed radiofrequency (2). These preliminarily positive results of more aggressive neuromodulatory interventions seem to indicate that the novel application of interventional approaches may hold promise in finding an effective long-term treatment for pudendal neuralgia.

SNS may be an interventional procedure to provide long-term relief. The main indications for SNS have included dysfunctions of the bladder and bowel, but it may have a role for the treatment of chronic pelvic pain (14,15). SNS was first approved for the treatment of urge incontinence in 1997 and for urinary frequency along with non-obstructive urinary retention in 1999 (16). Since that time, SNS has also been effective in treating patients with intractable pelvic pain with simultaneous urinary and fecal dysfunction (16-19). SNS has been used off-label in patients with interstitial cystitis and painful bladder syndrome and has been shown to improve their intractable pain (16,17,20,21). The use of SNS in the treatment of pudendal neuralgia as a cause of chronic pelvic pain, however, has not been described in the literature.

There are multiple approaches for placement of SNS electrodes. We employed the commonly used transforaminal approach, though it has an increased propensity for lead migration. Some physicians have begun to employ a caudal entry point or a cephalocaudal "retrograde" feeding of electrodes to the roots before they exit their respective foramina (22). The best approach for SNS placement for treatment of pudendal neuralgia should be determined by further studies.

The mechanism by which SNS can produce pain relief is not well understood, but the Gate Control Theory as first proposed by Melzack and Wall has long been a proposed explanation (23). Subsequent theories suggest that peripheral nerve stimulators may suppress dorsal horn activity or nociceptor axon firing (24-29). In our patient, SNS consisted of stimulation of the third and fourth sacral nerves bilaterally via quad electrodes (16). Sacral nerve stimulation by placement of implantable leads at the desired sacral level via a caudal epidural route is sometimes employed, but sacral nerve stimulator lead placement is more commonly achieved via an intraspinal, transforaminal, or extraforaminal approach (31-33). Historically, the extraforaminal technique has been used commonly and originally involved an invasive surgical exposure of the posterior aspect of the sacrum to facilitate lead placement into the S3 foramen under direct visualization (34). Extraforaminal SNS can now be done via a percutaneous minimally invasive approach under fluoroscopic guidance (35). The same technique has been further refined by neurophysiologic testing prior to permanent lead and implanted permanent generator (IPG) placement as used for SNS for fecal incontinence (14).

CONCLUSION

In summary, pudendal neuralgia is a difficult condition to diagnose and treat effectively. Based on research indicating SNS has had a positive effect in helping to relieve pelvic pain in patients with urinary and bowel dysfunction, we attempted to treat pudendal neuralgia by minimally invasive transforaminal SNS. This case appears to be the first report of isolated pudendal neuralgia successfully treated by transforaminal SNS. The patient continues to be more functional and has substantially less pain 4 years following the procedure. Therefore, transforaminal SNS appears to be a viable treatment option for isolated pelvic pain due to pudendal neuralgia refractory to conservative treatment. Larger scale studies should be undertaken to corroborate the effectiveness of this approach.

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