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# Sixty-Day Pudendal Nerve Stimulation: A Potential Therapy for Refractory Pudendal Neuralgia Case Report

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**Background:** Pudendal neuralgia (PN) can cause severe, disabling chronic pain. Though common, PN is frequently unrecognized and misdiagnosed. Historically, the last-resort treatment for PN has been permanent implantation of spinal cord stimulation (SCS), but SCS for PN carries high risk of complications and explantation. We report the first case of temporary (60-day) peripheral nerve stimulation (PNS) treatment for refractory PN.

**Case Report:** A 63-year-old woman presented with one year of chronic bilateral suprapubic vaginal pain radiating to the bilateral proximal medial thighs with concomitant dysuria, urinary frequency, and pain with intercourse. PN was confirmed via diagnostic pudendal nerve block. Using fluoroscopic guidance, we implanted PNS leads on the left and, subsequently, the right pudendal nerves, with explantation at 60 days for each lead. The patient reported continuing pain reduction with 80% improvement in the Visual Analog Scale score at 6 months, resumption of normal activity and functionality, discontinued use of opioids, and high satisfaction with treatment. This case is notable for the sustained pain relief provided by this temporary and minimally invasive treatment.

**Conclusions:** This case suggests that 60-day PNS with fluoroscopic guidance is a viable treatment for refractory PN in correctly selected patients. This treatment is low-risk, minimally invasive, and may be used early in the care continuum, potentially sparing patients multiple failed treatments and the risks associated with permanently implanted devices.

**Key words:** Pudendal nerve, peripheral nerve stimulation, neuromodulation, chronic pain, pelvic pain, perineal pain, case report

## BACKGROUND

Pudendal neuralgia (PN), which affects both women and men, is a chronic, debilitating form of perineal pain that is often refractory to conventional treatment (1). PN presents as piercing, unilateral, or bilateral neuropathic pain in the dermatome of the pudendal nerve (urogenital and/or anorectal) and is exacerbated by sitting (2). Women experience pain in the vulva, vagina, and clitoris, as well as posteriorly. For men, the urogenital pain is located in the testes, scrotum, and penis.

The true epidemiology of PN is unknown. While some have characterized PN as a rare disorder (3), the incidence of PN has been reported to be as high as 1% in the general population, with women experiencing the condition more frequently than men (4). PN is commonly misdiagnosed or unrecognized in clinical practice, and many practitioners who treat.

PN patients believe PN to be far more common than has often been reported in the literature (5). PN is as-

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sociated with significant impact on quality of life (6), including depression and anxiety, inability to maintain sexual relationship with partner, and relationship discord.

The first step in PN treatment is conservative management consisting of physical and pharmacological therapy. If pain continues to inhibit normal daily function, a pudendal nerve block is the first-line approach for both diagnosis and pain management (7). Psychological therapy (8) may also be part of an individualized, multimodal approach to treatment. Should these treatments fail, surgical decompression is often used for patients who respond to diagnostic nerve blocks. However, 30% of PN surgical decompression patients experience no pain relief (9). The last-resort treatment for intractable PN has been a trial of neuromodulation followed by permanent implantation of electrodes using fluoroscopic guidance. However, permanent implantation of neuromodulation devices (10) is an invasive treatment associated with risk of lead migration, breakage, and infection. Moreover, traditional spinal cord stimulation (SCS) in the pelvic region is anatomically challenging (7). In a retrospective analysis (11) of 243 patients treated for chronic non-cancer pain with permanent SCS implantation, abdominal/pelvic pain patients had the highest rate (33%) of explantation, most commonly due to loss of therapeutic effect. The rate of surgical site infection, another cause of explantation, was 4.3% among all patients. Based on limited evidence (12), dorsal root ganglion (DRG) stimulation may be more promising.

### Sixty-Day Stimulation of the Pudendal Nerve

A relatively new neuromodulation approach involves minimally invasive, 60-day percutaneous peripheral nerve stimulation (PNS) (13). PNS delivers electrical pulses through an implanted fine-wire lead to a targeted peripheral nerve. The lead is connected to a miniature wearable stimulator programmed by the clinician and adjusted by the patient to provide strong yet comfortable sensations. For patients with refractory neuropathic pain, PNS can serve as a test of the efficacy of permanently implanted neurostimulators; more importantly, it may deliver long-term pain relief without the need for further treatments. In a randomized, placebo-controlled trial of PNS for chronic postamputation pain (13), 67% of PNS patients reported ≥ 50% reductions in average weekly pain at 12 months compared with 0% in the control group at crossover. Similar 12-month results have been reported for PNS in chronic low back pain (14) and hemiplegic shoulder pain (15). Sixty-day PNS treatment (16) has also been demonstrated to reduce opioid use. To the best of our knowledge, this is the first reported case of 60-day PNS for refractory PN.

## CASE REPORT

This case presentation is institutional review board exempt per local institutional guidelines (45 CFR 164.501). The author obtained consent from the patient for the publication of the case. A 63-year-old woman presented with one year of chronic bilateral suprapubic vaginal pain with concomitant dysuria, urinary frequency, and pain with intercourse. The patient reported longstanding chronic low back pain, with one year of increasing paraspinal muscle pain and posterior superior iliac spine pain with radiation down both lateral thighs; findings from a previous magnetic resonance scan indicated disc herniation. The patient had previously visited an orthopedic surgeon who suggested her vaginal pain may be emanating from her herniated nucleus pulposus; the patient subsequently underwent epidural steroid injections without relief of her pelvic pain. Given the lack of response, the patient was then referred to a urogynecologist and underwent workup/treatment for suspected chronic urinary tract infection; this treatment was unsuccessful. Upon presentation to pain medicine, the patient prioritized treatment for her vaginal pain, which severely limited activities of daily living (ADLs), such as walking and driving. Pain medications included hydrocodone-acetaminophen 10/325 mg, mirabegron 25 mg (oral bladder relaxant), prasterone 6.5 mg (vaginal steroid suppository), progesterone 100 mg (oral hormonal therapy), and topical hormonal therapy via estradiol 0.05 mg and estradiol 0.025 mg patch. The patient completed 5 sessions of pelvic physical therapy (PT). The patient was utilizing hydrocodone/acetaminophen 10/325 mg 4 times per day, suggesting dependence. A computed tomography scan of her pelvis was unremarkable. A subsequent diagnostic local-only pudendal nerve block provided the patient short-term 100% improvement in her PN symptoms and confirmed a diagnosis of PN. Based on these findings, the patient chose to pursue bilateral 60-day PNS treatment for pain. Her baseline Visual Analog Scale score at the initial consult was 7/10 on average pain score; however, she reported 9/10 during ADLs, including driving, walking, and intercourse.

We implanted the first lead on her left pudendal

nerve and, one month later, the second lead on her right pudendal nerve, with removal of each lead 2 months after implant; therefore, the patient received simultaneous stimulation of both pudendal nerves for 30 days. Her pain quality was significantly improved with only mild intermittent pain, although still daily. Following removal of the initial (left) pudendal nerve lead, the patient reported continued pain reduction on her left side, but reduced benefit on her right side. We reprogrammed the PNS device to provide better coverage and performed troubleshooting on the device by switching out some external battery hardware; thereafter, the patient reported improvement on her right side. When the patient visited for removal of the second (right-side) lead, she reported 70% sustained improvement from baseline. Pain was located in the bilateral buttock with occasional shooting pain down the legs. At 6 months, the patient endorsed 80% bilateral sustained relief of vaginal pain; ability to drive, walk, and perform ADLs without pain; and even reported hiking in the Canadian Rockies. The patient also reported discontinued use of opioids for pain relief and was highly satisfied with her treatment. As the pudendal nerve has motor as well as sensory function, we inquired about any potential motor-related change in sensation, e.g., vaginal canal dilatation. The patient reported none.

## METHOD

For each of the 2 lead implantation procedures, the patient was brought into the procedure room and placed in the prone position on the fluoroscopy table. Standard monitors were placed and vital signs observed throughout the procedure. The left buttock area was prepped and draped in a sterile manner. The ischial spine was identified with anterior/posterior fluoroscopy with a 15° caudal and 10° ipsilateral oblique tilt (Fig. 1). The skin and subcutaneous tissues in the area were anesthetized with 1% lidocaine. A 20-guage stylet cannula was advanced toward the ischial spine under fluoroscopic guidance until the bone was contacted. The needle was then walked off the bone in an inferomedial fashion. Initial aspiration was negative for venous blood. One mL of contrast dye was easily injected via a 20 g angiocatheter attached to a 5 mL syringe and showed appropriate spread; absence of piriformis muscle and sciatogram were noted (Fig. 2). An image representing piriformis uptake may distort results as it activates an unwanted muscle sensation (Fig. 3). The cannula was connected to the device for testing, which demonstrated paresthesia over the affected area. Leaving the cannula in place, the stylet/wire was removed and replaced with the implantable lead (MicroLead; SPR Therapeutics). Subsequent testing continued to demonstrate paresthesia over the affected area. The cannula and stylet were removed, leaving the implantable lead in place. The lead was connected to the miniature wearable stimulator (SPRINT PNS System; SPR Therapeutics) and continued to provide paresthesia over the affected area. The cannula and stylet were removed, leaving the lead in place. The lead was secured with Dermabond® and dressings, and the area was infiltrated with 1% lidocaine. The patient's back was cleaned and dressed. The patient tolerated the procedures well, with stable vital signs and no apparent complications. After each procedure, the patient was taken to the recovery area where written discharge instructions were given. The patient was instructed to maintain light activity for one week following each procedure to allow for stabilization of the implanted leads. After implantation of the first lead, the procedure was repeated on the contralateral side 30 days later.

## DISCUSSION

To the best of our knowledge, this is the first reported case of 60-day PNS for treatment of PN. The case is notable for the sustained pain relief provided by this temporary and minimally invasive treatment. We highlight this case as a potential minimally invasive treatment option for a painful and emotionally disabling condition in both women and men that has historically lacked successful treatment options.

PN is a particularly cruel disorder, marked by relentless pain that has been compared to an acute toothache (2). Patients typically see a number of specialists and endure several unsuccessful treatments before the pudendal nerve is identified as a treatment pathway. Thus, they endure prolonged chronic pain (17) that may heighten risk of maladaptive structural plasticity in neural circuits and perpetuate pain chronicity (18). Neuromodulation (19) has been described as a "digital drug" for chronic pain to counter opioid use. The potential attraction of 60-day PNS is that it offers a low-risk, minimally invasive treatment option early in the treatment continuum. At a minimum, this treatment serves as a trial for permanently implanted neuromodulation devices should pain return after removal of leads. Better still, pain relief may be sustained without further, more invasive treatments (13-15). Moreover, 60-day PNS may be especially useful as a neuromodulation option for PN. Whereas, the sacral nerve

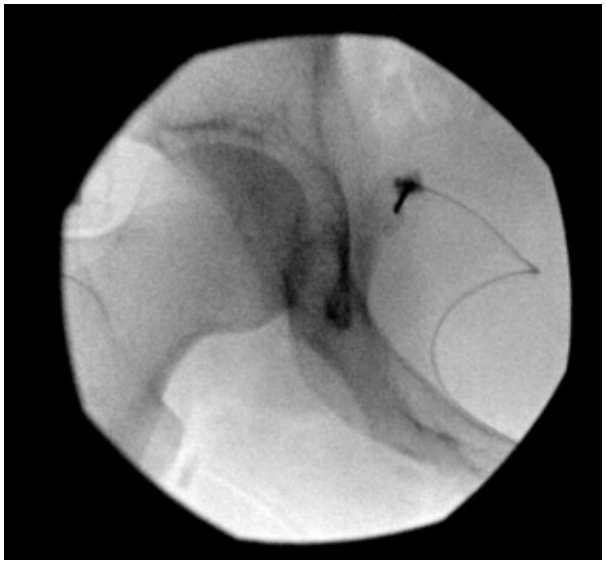

Fig. 1. Appropriate location of needle utilizing ipsilateral and caudal tilt to identify the ischial spine. The target location is the "apex" of the triangular-shaped ischial spine.

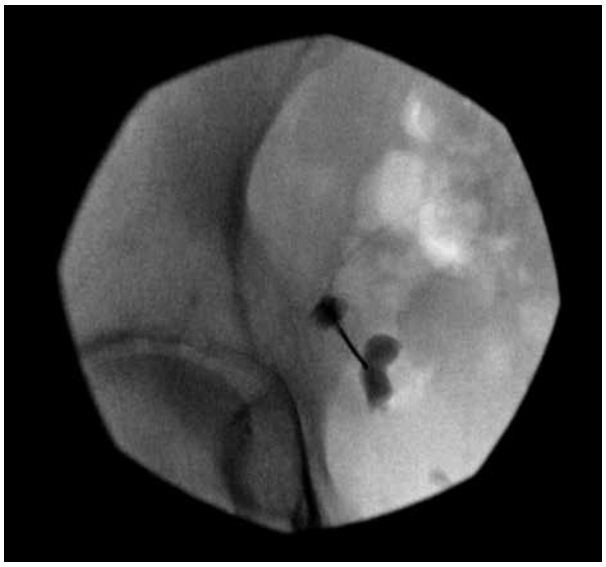

Fig. 2. Omnipaque 240 mgi/mL contrast spread indicating appropriate spread without piriformis and sciatic nerve uptake.

roots are notoriously difficult to recruit for traditional SCS (7), PNS allows for straightforward access to the pudendal nerve just as it exits the sacral canal. In our clinic, patient selection criteria for 60-day PNS includes pelvic pain over the pudendal nerve distribution, unsuccessful trial of medical management and pelvic PT, and response to an Abdi-technique (20) diagnostic nerve block.

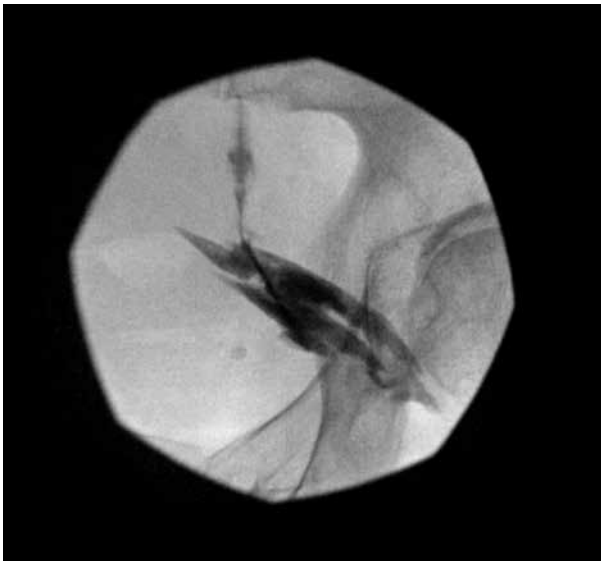

Fig. 3. Contrast spread demonstrating piriformis uptake. This illustrates the importance of using contrast during this procedure to avoid unwanted and painful piriformis involvement and stimulation

## CONCLUSIONS

Though ultrasound guidance is often used for PNS lead placement in other settings, we use fluoroscopic guidance for lead placement on the pudendal nerve. As the preferred technique for pudendal nerve blocks, fluoroscopic guidance is already familiar to PN practitioners. Furthermore, it allows for use of contrast to ensure the piriformis muscle and sciatic nerve are avoided. Depending on adipose deposition on the buttocks, a pudendal nerve block may be as deep as 6-7 cm, which may limit identification of important structures via ultrasound.

As noted, this patient experienced reduced pain relief on her right side when we removed the left-side lead after 30 days of dual left- and right-side stimulation. Her right side improved when we reprogrammed the device. We speculate that the initial reduction in right-side pain relief may have been due to loss of stimulation from the left side. The left and right branches of the pudendal nerve are in close proximity (~1 mm) and have interconnecting fibers that may have facilitated some carryover of electric current.

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## REFERENCES


1. Popeney C, Ansell V, Renney K. Pudendal entrapment as an etiology of chronic perineal pain: Diagnosis and treatment. *Neurourol Urodyn* 2007; 26:820-827.
2. Robert R, Prat-Pradal D, Labat J, et al. Anatomic basis of chronic perineal pain: Role of the pudendal nerve. *Surg Radiol Anat* 1998; 20:93-98.
3. Stav K, Dwyer PL, Roberts L. Pudendal neuralgia: Fact or fiction? *Obstet Gynecol Surv* 2009; 64:190-199.
4. Spinosa J-P, De Bisschop E, Laurençon J, Kuhn G, Dubuisson J, Riederer BM. Sacral staged reflexes to localize the pudendal compression: An anatomical validation of the concept. *Rev Med Suisse* 2006; 2:2416-2418, 2420.
5. Hibner M, Desai N, Robertson LJ, Nour M. Pudendal neuralgia. *J Minim Invasive Gynecol* 2010; 17:148-153.
6. Health Organization for Pudendal Education. Personal stories. Accessed Nov. 22, 2020. www.pudendalhope.info/node/17
7. Hunter CW, Stovall B, Chen G, Carlson J, Levy R. Anatomy, pathophysiology and interventional therapies for chronic pelvic pain: A review. *Pain Physician* 2018; 21:147-167.
8. Gatchel RJ, Peng YB, Peters ML, Fuchs PN, Turk DC. The biopsychosocial approach to chronic pain: scientific advances and future directions. *Psychol Bull* 2007; 133:581-624.
9. Robert R, Labat J-J, Bensignor M, et al. Decompression and transposition of the pudendal nerve in pudendal neuralgia: A randomized controlled trial and long-term evaluation. *Eur Urol* 2005; 47:403-408.
10. Vakkala M, Järvimäki V, Kautiainen H, Haanpää M, Alahuhta S. Incidence and predictive factors of spinal cord stimulation treatment after lumbar spine surgery. *J Pain Res* 2017; 10:2405-2411.
11. Hayek SM, Veizi E, Hanes M. Treatment-limiting complications of percutaneous spinal cord stimulator implants: A review of eight years of experience from an academic center database. *Neuromodulation* 2015; 18:603-609.
12. Hunter CW, Yang A. Dorsal root ganglion stimulation for chronic pelvic pain: A case series and technical report on a novel lead configuration. *Neuromodulation* 2019; 22:87-95.
13. Gilmore C, Ilfeld B, Rosenow J, et al. Percutaneous 60-day peripheral nerve stimulation implant provides sustained relief of chronic pain following amputation: 12-month follow-up of a randomized, double-blind, placebo-controlled trial. *Reg Anesth Pain Med* 2020; 45:44-51.
14. Gilmore CA, Kapural L, McGee MJ, Boggs JW. Percutaneous peripheral nerve stimulation for chronic low back pain: Prospective case series with 1 year of sustained relief following short-term implant. *Pain Pract* 2020; 20:310-320.
15. Chae J, Yu DT, Walker ME, et al. Intramuscular electrical stimulation for hemiplegic shoulder pain: A 12-month follow-up of a multiple-center, randomized clinical trial. *Am J Phys Med Rehabil* 2005; 84:832-842.
16. Ilfeld BM, Ball ST, Cohen SP, et al. Percutaneous peripheral nerve stimulation to control postoperative pain, decrease opioid use, and accelerate functional recovery following orthopedic trauma. *Mil Med* 2019; 184 (suppl 1):557-564.
17. Kuner R, Flor H. Structural plasticity and reorganisation in chronic pain. *Nat Rev Neurosci* 2017; 18:20-30.
18. Latremoliere A, Woolf CJ. Central sensitization: A generator of pain hypersensitivity by central neural plasticity. *J Pain* 2009; 10:895-926.
19. Deer TR. Neurostimulation should be used as a method of reducing or eliminating opioids in the treatment of chronic pain: The digital drug revolution. *Expert Rev Med Devices* 2013; 10:697-699.
20. Abdi S, Shanouda P, Patel N, Saini B, Bharat Y, Calvillo O. A novel technique for pudendal nerve block. *Pain Physician* 2004; 7:319-322.