

Case Report

 **Pudendal Entrapment Neuropathy: A Rare Complication of Pelvic Radiation Therapy**

Foad Elahi, MD, Daniell Callahan, MD, Jeremy Greenlee, MD, and Tammy L. Dann, MD

From: University of Iowa Hospital, Iowa City, Iowa

Dr. Elahi is a Clinical Assistant Professor, Department of Anesthesia, University of Iowa Hospital, Iowa City, Iowa. Dr. Callahan and Dr. Greenlee are Assistant Professors at the University of Iowa, Iowa City, Iowa. Dr. Dann is a Pain Fellow at the University of Iowa, Iowa City, Iowa.

Address Correspondence:  
Foad Elahi, MD  
200 Hawkins Dr  
University of Iowa  
Iowa City, Iowa  
E-mail:  
foad-elahi@uiowa.edu

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Pudendal nerve entrapment (PNE) is an uncommon cause of chronic pain. Pudendal nerve entrapment typically occurs when the pudendal nerve is fused to nearby anatomical structures or trapped between the sacrotuberous and sacrospinous ligaments. Pudendal nerve entrapment can be caused by excessive bicycling, pregnancy, anatomic abnormalities, scarring due to surgery, or as a sequela of radiation therapy. Radiation-induced peripheral neuropathy is usually chronic, progressive, and often irreversible. Radiation-induced pudendal neuropathy is much less common than the more familiar brachial plexopathy secondary to radiation treatment for breast cancer. The prevalence of PNE, however, is increasing due to improved long-term cancer survival.

Diagnosis of pudendal neuralgia is essentially clinical; no specific clinical signs or complementary tests are reliably confirmatory.

A detailed pain history with correlative clinical examination is paramount for accurate diagnosis. Performance of a pudendal nerve block can serve as both a diagnostic and therapeutic tool.

Utilization of various imaging studies, as well as the performance of an electrophysiological study with pudendal nerve motor latency testing, may yield valuable evidence in support of a pudendal neuralgia diagnosis.

We present the case of a 59-year-old man with stage IV prostate cancer, referred to the pain clinic for chronic perineal and right sided pelvic pain. His pain began insidiously, approximately 2 months after undergoing radiation treatment and chemotherapy 3 years prior. He was ultimately diagnosed as having a right sided pudendal entrapment neuropathy. His pain was refractory to all conventional treatment modalities; therefore we decided to pursue neuromodulation via a dorsal column spinal cord stimulator implant. Below, we describe the decision making process for the diagnosis and treatment of his pudendal neuropathy.

**Key words:** Pudendal nerve entrapment, neuralgia, neuromodulation, spinal cord stimulation.

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**T**he pelvis is a structure that presents itself to the attention of multiple medical specialists. To ascertain the etiology of chronic pelvic pain is often a lengthy process which sometimes yields fruitless results. The pudendal nerve arises from the sacral plexus and is formed by the second, third, and fourth sacral

nerve roots. The nerve exits through the greater sciatic foramen, crossing the ischial spine, the sacrospinous ligament, and the sacrotuberous ligaments (1).

Pudendal neuropathy is a rare affliction whose diagnosis is elusive; patients often end up having inappropriate and unnecessary surgeries. [NOTE: Pudendal

neuralgia is a symptom and pudendal neuropathy is a diagnosis.]

There are 2 main entrapment locations, eventually associated: in the claw between the sacrospinous and sacrotuberous ligaments near the ischial spine and in the Alcock canal due to the falciform process of the sacrotuberous ligament and/or fascia of the obturator internus muscle.

Pudendal neuropathy is essentially a clinical diagnosis; no specific clinical signs or complement of tests are reliable (2). A detailed pain history and physical exam is essential. Pudendal neuropathy secondary to nerve entrapment causes neuropathic pain with varying degrees of pain intensity in the S3-S4 dermatome. The nerve itself may become entrapped at several points along its course leading to intractable pelvic and perineal hyperalgesia. Other symptoms may include genital numbness and sexual dysfunction that can confound the diagnosis.

Pudendal nerve entrapment may result from trauma, infection, or malignancy and often presents after childbirth in women. The clinical diagnosis is based on the Nantes criteria (3).

A physical exam is not confirmatory in many cases. Reproducing the pain during palpation of the rectum in the ischial spine region may or may not occur. Due to the chronicity of pain and the psychologically sensitive pelvic region, the perineum and genitalia may have a decreased threshold response to tactile stimuli. Pelvic muscle deconditioning, especially in the deep pelvic muscles like the piriformis muscle, may contribute to the pain. Chronic pelvic pain patients commonly report coccygeal pain, genital hypersensitivity, and other painful conditions that can also confound a diagnosis. An interventional pain physician should be consulted when a more conventional diagnosis such as prostatitis, vaginitis, or urinary tract infections have been ruled out.

## **METHODS**

Radiographic and electrodiagnostic tools such as pelvic x-ray, magnetic resonance imaging (MRI), magnetic resonance neurography, computerized tomography, and pudendal nerve motor latency test are utilized to support the diagnosis (3).

A pudendal nerve block using a local anesthetic can be diagnostic, as well as therapeutic if combined with steroids. A transvaginal, transrectal, or posterior approach to reach the pudendal nerve at the ischial spine where it is most often entrapped between the sacrospinous and sacrotuberous ligaments can be ac-

complished with the help of an ultrasound machine.

The injection can also be given "blindly" (without imaging), however, noninvasive imaging modalities provide improved safety and efficacy since accurate placement of the needle is paramount in confirming a diagnosis.

Surgical nerve decompression becomes an option if injections fail to provide an adequate duration of relief. With more recent developments focusing on less invasive treatment modalities, multimodal pain management and neuromodulation have become popular for treating chronic neuropathic pain.

## **CASE PRESENTATION**

We present the case of a 59-year-old man diagnosed with stage IV prostate cancer who had a Gleason score of 9. He received chemotherapy and external beam radiation therapy that was mapped based upon regional lymph node involvement and MRI findings. The total doses to the entire pelvic region, including pelvic lymph nodes and the prostatic bed, was approximately 74 Gy in a conventional fractionation.

At one-year follow-up, pelvic MRI and nuclear bone scans were negative for a tumor. In the absence of local disease, no metastasis, and negative repeat biopsies, his oncologist considered the treatment successful.

Approximately 2 months after completing chemotherapy and radiation therapy, he developed an insidious onset of perineal and pelvic pain. His pain was primarily in the left pelvic and perineal region with intermittent radiation to the testicles and the left groin. He described the pain as deep, aching, and sharp with an occasional burning quality. Sitting for a prolonged period of time greatly exacerbated the pain. Oral pain medications, including high doses of gabapentin, amitriptyline, and morphine failed to provide adequate pain relief.

He was seen by other pain physicians and underwent a caudal epidural steroid injection, pelvic trigger point injections, a ganglion impar block, and pelvic floor physical therapy, all of which were unsuccessful.

A left pudendal nerve electrodiagnostic study showed an increase in the distal motor latency of the distal motor branch of the pudendal nerve. Lumbosacral and pelvic x-ray and MRI were unrevealing.

After a thorough evaluation and review of previous imaging, we decided to perform a left pudendal nerve block under ultrasound guidance.

The patient was placed in the prone position. Under sterile conditions with proper preparation and

draping, the perineal region was explored with a curved ultrasound probe. The ischial spine, sacrotuberous ligament, and the internal pudendal artery were delineated by ultrasound (Fig. 1). The pudendal nerve accompanies the pudendal artery.

A 22-gauge 3.5-inch spinal needle was inserted via the in-plane technique. The needle trajectory was guided under ultrasound guidance. After the needle tip was visualized at the targeted area, 5 mL of normal saline was injected and utilized as hydro-dissection to clearly identify the spread of the subsequent injectate. We observed an increase in the distances between sacrospinous and sacrotuberous ligaments. After negative aspiration for blood, 3 mL of 2% lidocaine was injected. The patient tolerated the procedure well and reported his pain score decreased from 8 to 2 (numeric rating scale) (Fig. 2).

In order to rule out the placebo effect from the initial nerve block, a second confirmatory nerve block was later performed using 3 mL of bupivacaine 0.5% which has a different duration of action. The second block also provided greater than 80% pain relief and the duration was concordant with the local anesthetic used. These observations were consistent with a diagnosis of pudendal entrapment neuropathy.

At this point, we decided to proceed to a trial of dorsal column neuromodulation. Under fluoroscopic view, the thoracolumbar region was prepped with povidone-iodine and a full body drape was applied. Using complete aseptic technique, the L1-L2 intervertebral space was identified with the help of fluoroscopy. Lidocaine 1% was used for skin and needle tract infiltration. A 15-gauge Tuohy needle was advanced under fluoroscopic guidance and placed in the epidural space between L1-L2. The needle position was confirmed on anterior, posterior, lateral, and oblique views. At that point, an 8-contact lead (Medtronic 1x8 Standard External Trialing Neurostimulator Model 3777, Medtronic, Minneapolis, MN) was inserted with electrode 0 positioned at the top of the T9 vertebra and the lowest lead positioned on the lower border of the T10 vertebra. The lead was connected to a pulse generator and various combinations of electrodes, amplitudes, and pulse widths were utilized to create paresthesia in the distribution of the patient's pain. The patient tolerated the spinal cord stimulator (SCS) trial procedure without complication. Subsequently, over the next 7 days, he used neurostimulation 100% of the time. He reported very good pain relief and was very satisfied with the trial of SCS. Overall, his pain decreased from a pretrial

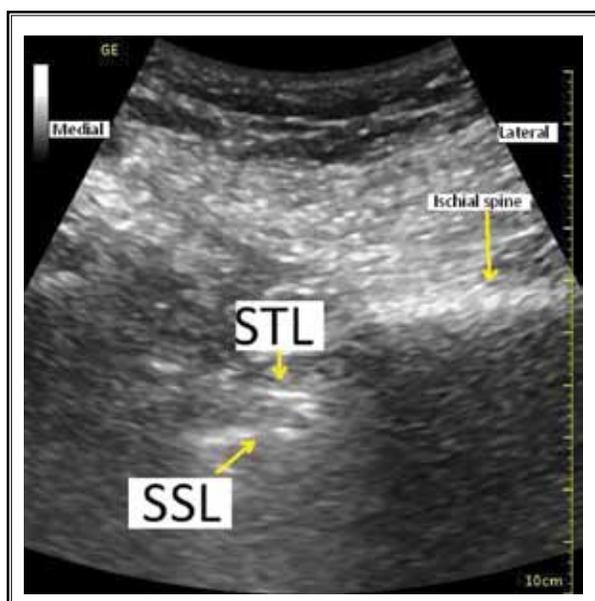


Fig. 1. The sacrospinous ligament and sacrotuberous ligament were delineated by ultrasound.

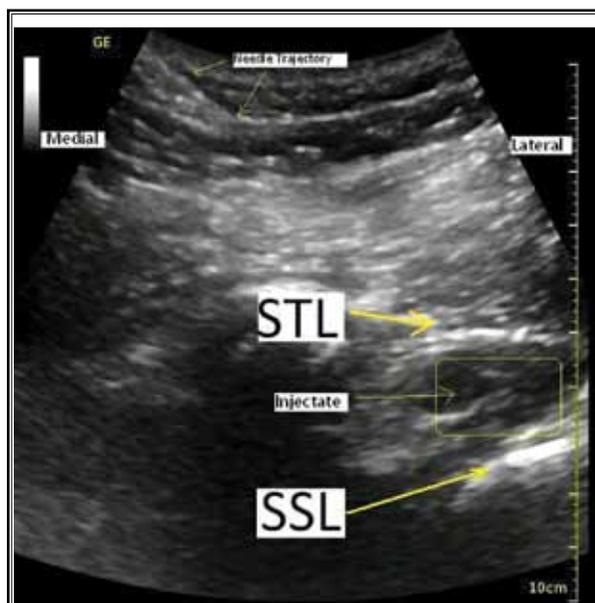


Fig. 2. Increase in the distances between sacrospinous ligament and sacrotuberous ligament after injection.

score of 8 to 1 with the spinal cord stimulator. He also was able to decrease his medication usage during the trial. Based on his response to the SCS trial, the patient underwent permanent implantation. A 16-electrode

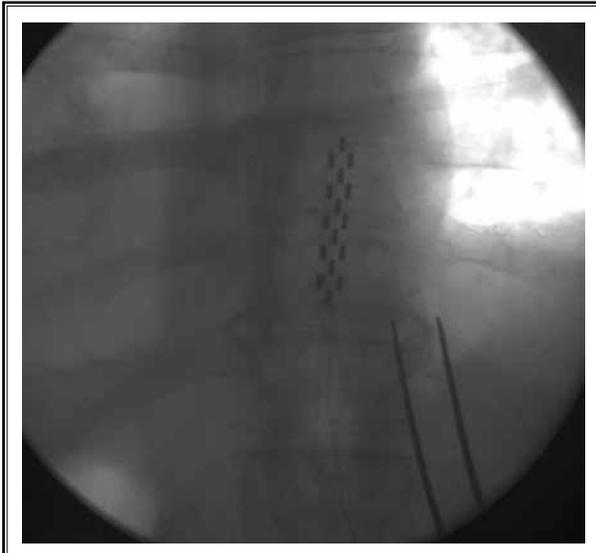


Fig. 3. Location of the dorsal spinal cord permanent implantation at the T9-T10 level.

Medtronic Surgical Lead (5-6-5 Model 39565) was implanted (Fig. 3).

At his follow up appointment 10 months after the surgery, he reported 100% overall satisfaction with treatment and a numerical pain score of 1-2 for perineal pain. He was also able to discontinue gabapentin and his use of opioid medications. He continues to take amitriptyline 25 mg at night and acetaminophen as needed.

## Discussion

The goal of radiation therapy for patients with localized prostate cancer is the delivery of a tumoricidal dose of radiation while minimizing radiation to surrounding normal tissues. Unfortunately, pudendal neuropathy can be a sequela of radiation therapy.

Radiation-induced peripheral neuropathy may present as a static clinical presentation or become progressively worse. It may appear several months to several years after radiation treatment is completed. Its occurrence is rare but increasing with improved long-term cancer survival. There is great clinical heterogeneity in the neurological presentation since various anatomic sites are irradiated. Radiation-induced neuropathy is often irreversible and refractory to most medical management.

The pathophysiological mechanisms proposed in the medical literature include nerve compression by indirect extensive radiation-induced fibrosis, direct injury to nerves, and injury to blood vessels by ischemia (4).

In this case report, we observed a pudendal neuralgia that had a clear association with pelvic irradiation. Based on our best knowledge, this is a unique case report in the medical literature that shows a clear relationship between radiation and neuralgia in the distribution of the pudendal nerve.

His pain was refractory to medication management including antiepileptics, antidepressants, muscle relaxers, opioids, caudal epidural injection, ganglion impar block, biofeedback, coping, hypnotherapy, and relaxation therapy.

Electrical spinal cord modulation (dorsal spinal cord stimulation) is commonly used for various neuropathic pain syndromes including but not limited to failed back surgery syndrome, complex regional pain syndrome, neuropathic pain, angina, and ischemic limb pain. The spinal cord has proved to be an excellent target for treatment in many types of chronic neuropathic pain. Despite a plethora of publications postulating the mechanism of spinal cord stimulation, the precise mechanism of action remains unclear. Spinal cord stimulation has been well documented to relieve chronic intractable neuropathic pain (5).

Successful application of minimally invasive techniques of spinal cord stimulation as an alternative to surgical intervention may have profound implications for patients suffering from pelvic and perineal pain.

Percutaneous implantation of small profile leads in some patients may be preferable over surgical implanted paddle leads that need a small laminectomy with or without general anesthesia.

Other modalities for the treatment of pudendal neuralgia besides surgical pudendal nerve decompression that allow all possible entrapments to be corrected are described in the medical literature, including the utility of a pudendal nerve block by conventional or pulsed radiofrequency ablation and cryo nerve ablation.

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