



Pudendal neuralgia, a severe pain syndrome

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Received for publication April 21, 2004; revised December 28, 2004; accepted January 20, 2005

KEY WORDS

Pudendal neuralgia
Electrodiagnosis
Pudendal injection
Pudendal
decompression
Pudendal
neuromodulation
Outcomes

Objective: To describe the clinical and electrodiagnostic findings, therapies, and outcomes of patients with pudendal neuralgia.

Study design: A retrospective, descriptive study of 64 patients from March 19 to December 22, 2003.

Results: Clinical findings included pain along nerve distribution (64, 100%), pain aggravated by sitting (62, 97%), pain relieved by standing or lying (57, 89%), and misdiagnosis (53, 83%). Neurophysiologic findings were normal (23, 35%), demyelination (17, 26%), axonal loss (5, 7.5%), and demyelination with axonal loss (21, 32%). Therapies were conservative (64, 100%), nerve injection (38, 59%), neuromodulation (2, 3%), and decompression surgery (10, 15%). Slight or moderate pain improvement with therapies included conservative (64, 100%), nerve injection (12, 31%), neuromodulation (2, 100%), and decompression (6, 60%).

Conclusion: Pudendal neuralgia is poorly recognized and poorly treated. Improvement is gained with conservative therapy. Injections and decompression benefit one half and one third of patients, respectively. Neuromodulation needs further evaluation.

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In this day of patient use of Web sites, a few centers with focus on caring for patients with pudendal neuralgia have been recognized, leading to an inordinate number of patient encounters by those centers.

Neuralgia is defined as “severe, sharp pain along the course of a nerve.”¹ When the pain involves the sensory distribution course of the pudendal nerve, the term “pudendal neuralgia” is appropriate.

The pudendal nerve sensory distribution course is generally via the 3 terminal branches; inferior rectal,

perineal, and dorsal nerve of the penis/clitoris. The inferior rectal supplies the integument around the anus and communicates with the perineal branch of the posterior femoral cutaneous nerve and its terminal branch, the posterior scrotal (labia majora) nerve. The perineal branch has a deep motor portion and 2 superficial sensory branches, the medial and lateral posterior scrotal (labial) nerves. The dorsal nerve of the penis/clitoris runs along the dorsum of the penis/clitoris, supplying the overlying skin. Pudendal neuralgia may present with pain in the distribution of some or all of these branches.

We review our experience with 64 patients presenting with pudendal neuralgia from March 19 to December 22, 2003. Clinical and electrodiagnostic

Presented at the Joint Scientific Meeting of the Society of Gynecologic Surgeons and the American Urogynecologic Society, July 29–31, 2004, San Diego, Calif.

Reprints not available from the authors.

Table Directed questionnaire

I have had a pain problem for how long? _____		
Is there an event that coincided with the original onset of the pain? _____		
The pain, on a scale with 0 being none and 10 being the worst possible is: _____		
The pain is (circle): "burning," "pulling," "crushing," "pressure," "throbbing," knife-like," "other," in nature.		
T	F	The pain is more on one side of my bottom than the other.
T	F	I have pain in the buttock region.
T	F	I have pain in the vagina/penis.
T	F	I have pain in the labia/scrotum.
T	F	The pain is worse with sitting.
T	F	Sitting on a commode is not as painful as sitting on a flat surface.
T	F	The pain is worse with standing.
T	F	The pain is worse lying down.
T	F	The pain is worse lying on one side than the other.
T	F	There is pain with bowel movement.
T	F	There is pain with intercourse.
T	F	There is pain with urinating.
T	F	I have trouble with bladder control.
T	F	I have trouble with bowel control.
T	F	I have trouble controlling bowel gas.

findings, therapies, and outcomes are summarized to increase awareness and advance therapy for patients with this condition.

Material and methods

All patients presenting with the complaint of pain in the distribution of the pudendal nerve, from March 19 to December 22, 2003, had systematic review of charts to record clinical and electrodiagnostic findings, therapies, and outcomes. This study was exempted from the institutional review board because all information was extrapolated from patients' charts, and no individual patient identification was directly or indirectly made.

Evaluation included a directed questionnaire (Table) and physical examination, and electrodiagnostic testing. Electrodiagnostic testing included pudendal nerve terminal motor latencies, electromyography of the right and left sphincter ani, and the bulbocavernosus reflex.²

An electrodiagnostic examination confirming the clinical impression is reassuring for the patient and physician; gives objective evidence of the severity of the condition; is helpful in planning treatment; and will be useful in understanding new, changed, or recurrent symptoms later in the patient's life.³

Therapies included medical management, pudendal nerve injections, pudendal decompression surgery, and pudendal neuromodulation.

Conservative management utilized both sitting on pads and drug therapy. The pads were specially designed with a cutout to allow sitting on the ischial tuberosities and preventing pressure on the perineum. Patients were also counseled to avoid postures that flex the thigh and

place potentially stretching forces on the pudendal nerve. Medications, which had been tried with varying success prior to our consultation included gabapentin, amitriptyline, nonsteroidal anti-inflammatories, skeletal muscle relaxants, and opioid analgesics. Modifications in dosage were made when appropriate.

Pudendal nerve injections were initially attempted under guidance with computed tomography,⁴ which allows anatomical localization. However, the response is variable and it requires scheduling through radiology. Anatomy review determined the pudendal nerve was accessible through a posterior approach, which allows office performance of injections. The patient is prone. A commercially available sponge electrode is placed in the vagina (females) or rectum (males). A portable electromyography (EMG) unit is used for stimulation and recording. The ischial tuberosity and the falciform process are palpated on a horizontal line at the level of the midanus (Figure). The area is cleansed. A 5-inch foramen needle is inserted medial to the sacrotuberous ligament at the level of the midanal line and directed into and through the ischioanal fossa toward the ipsilateral ischial spine. A micrograbber is used to stimulate through the foramen needle while recording from the sponge electrode. Simultaneously, the oscilloscope is monitored for compound muscle action potentials from the pelvic floor, biological response, contraction of the external anal sphincter, and reproduction of the patient's pain symptom. Once appropriate responses are obtained, a syringe of 9 cc 1% lidocaine and 1 cc triamcinolone is attached to the foramen needle.

Aspiration is performed to detect and prevent vascular placement and the medications are injected. Within

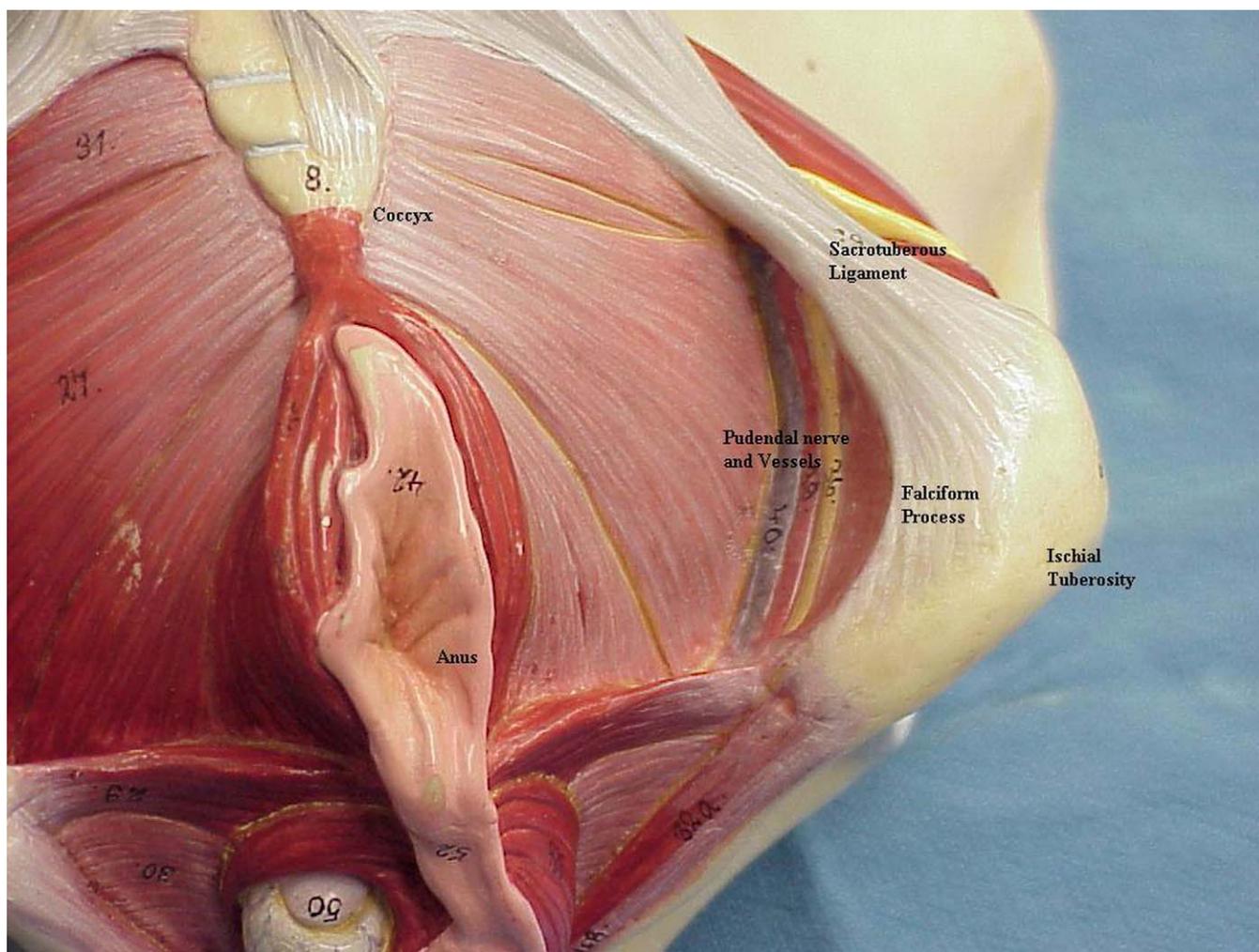


Figure Pudendal anatomy, posterior approach.

minutes there will be hypesthesia along the pudendal nerve distribution. It is important to monitor for muscle contraction in the legs while performing the injection. If noted, the foramen needle should be repositioned to obtain responses without leg involvement. This prevents injection of the lidocaine in the region of the sciatic nerve, which may cause temporary weakness of the lower limb. Patients are asked to have someone available to drive them home. The hypesthesia will last for a few hours. About 50% of patients report an exacerbation of symptoms over the subsequent 7-10 days. Relief is usually evident within 2 weeks. Duration of pain improvement, if achieved, is variable, ranging from a week to months.

To prevent possible hypothalamic-pituitary-adrenal suppression, the injections should not be repeated more frequently than every 6 weeks. Monitoring with a pain scale can be useful and should be considered early in the management course and as part of any study protocol. Subjects who did not receive relief from the initial injection received repeat injections at 6-week intervals up to 3 injections.

Patients with electrophysiologic evidence of pudendal neuropathy with axonal loss were offered pudendal nerve decompression. Pudendal nerve decompression surgery was performed as follows. The patient has general anesthesia without long-acting neuromuscular blockers. Positioning, EMG, and anatomy are as described above for injections. A line is drawn from the ischial tuberosity to the sacrum along the course of the sacrotuberous ligament. The patient is prepped in sterile fashion. It is important to make the incision and carry the dissection directly on top of the sacrotuberous ligament without straying medial or lateral. The pudendal neurovascular bundle runs between the sacrotuberous and sacrospinous ligaments. A Babcock clamp is useful to surround and elevate the sacrotuberous ligament while incising it at the falciform process. Incision is done with electrocautery until there a few millimeters left and is completed with scissors. The divided sacrotuberous ligament is elevated and retracted with a Kocher clamp. Then the neurovascular bundle can be visualized. However, there can be multiple structures in

this area that resemble nerve but are not. We use a blunt introducer needle, which allows stimulation with a micrograbber and monitor the EMG oscilloscope for compound muscle action potentials and the external anal sphincter for contraction. By identifying nerve activity, we are able to dissect and free the nerve safely as it passes through the ischioanal fossa and Alcock's canal.

Electrical stimulation of nerves to treat pain syndromes is well known, and the mechanism of action, "gate theory," whereby stimulation of large-diameter somatic fibers impairs impulse delivery by small fibers to higher centers has been described previously.⁵ Combining the techniques of pudendal nerve injection and sacral neuromodulation a tined quadripolar lead is placed by the pudendal nerve in the ischioanal fossa. A pilot study has demonstrated the feasibility and safety of pudendal neuromodulation for overactive bladder.⁶ An external generator is used to test the lead for a minimum of 2 to 4 weeks. If patients felt there was significant pain relief, the quadripolar lead is connected to an implanted pulse generator at a second-stage procedure.

Results

Sixty-four patients were studied: 18 males and 46 females, ranging in age from 30 to 71 years. The hallmark of pudendal neuralgia is the symptom of pain in the pudendal nerve distribution, which is aggravated by sitting. In 57 (89%) patients, pain was less with sitting on a commode and was relieved by standing or lying. It is important to note that 55 patients (86%) were diagnosed and treated for other conditions prior to a correct diagnosis of pudendal neuralgia. These patients had seen other primary care and specialty physicians. Some were treated for pain without a specific diagnosis, whereas others were diagnosed with other conditions.

Pudendal nerve clinical neurophysiologic findings were normal in 23 (35%), demyelination in 17 (26%), axonal loss in 5 (7.5%), and demyelination with axonal loss in 21 (32%).

Therapies were conservative in 100%, nerve injection in 38 (59%), pudendal nerve stimulation in 2 (3%), and decompression surgery in 10 (15%).

Pain improvement occurred slightly or moderately in all with conservative therapy, 12 (31%) with nerve injection, both patients with nerve stimulation, and 6 patients (60%) with surgical decompression.

Comment

The etiologies of neuralgias generally are considered to be nerve trunk compression, faulty nutrition, toxins, and inflammation. The anatomical course of the pudendal

nerve makes compression a likely factor, with the majority of cases of pudendal neuralgia.

The pudendal nerve arises from sacral nerves 2, 3, and 4 and passes in close association with the sciatic nerve between the piriformis and coccygeus muscles. The nerve crosses the ischial spine as it first leaves and then re-enters the pelvis through the greater and lesser sciatic foramina, respectively. At the area of the ischial spine, it is superficial to the sacrospinous ligament and deep to the sacrotuberous ligament, an area of possible compression or fixation. The nerve then accompanies the internal pudendal vessels along the lateral wall of the ischioanal fossa in a tunnel formed by a splitting of the obturator fascia (Alcock's canal). This tunnel is another area of possible compression or fixation. This portion of the obturator fascia is thickened and fibrous, and it is attached in part to the falciform process of the sacrotuberous ligament. The falciform (*L. falx*, sickle, +*forma*, form, sickle-shaped) process is formed by the dense, fanning-out bands of the medial portion of the sacrotuberous ligament as it attaches to the ischial tuberosity.

The anchoring of the obturator fascia, with its investing of the pudendal nerve to the falciform process forms another site of pudendal nerve mobility restriction. Upward forces on the perineum may then place stretching forces on the nerve to create localized compression of the nerve at a restriction site. The branching of the pudendal nerve during its course is variable. Thus, compression at any of the above sites may involve variable branches of the nerve. In addition, because the compressive pudendal nerve injuries lead to partial, rather than total nerve interruption, clinical manifestations are variable secondary to the topographical arrangement of the nerve's fascicular composition.⁷

When compressed, as by an incompatibility of volume of nerve structure and available anatomic space or by stretch of a nerve restricted in its motion, the biological effects are related to ischemia.⁸ In acute situations, the compressive effects of the ischemia lead to loss of conduction, particularly in the slower conducting (smaller) myelinated fibers. Such conduction block is associated with ectopic impulses generated along the nerve with perceived paresthesias.

There is no structural nerve damage and recovery is rapid if the acute compression is relieved. Situations with acute compression causing pain may be termed *functional entrapments*. In these situations, the pain is limited to the acute situation associated with the compressing maneuver (eg, sitting) with pudendal neuralgia. Other examples of functional entrapments are some cases of thoracic outlet syndrome, pronator syndrome, supinator syndrome, medial epicondylitis, and piriformis syndrome. Electrodiagnostic tests are generally normal because they are not performed in the aggravated entrapping position.

Peripheral nerves respond to chronic compression by an inflammatory reaction including venous stasis, increased vascular permeability, edema, and scar formation. Relatively early in the course of chronic compression, sustained conduction block with paranodal demyelination occurs. The internodes at the edges of the compressive zone are particularly distorted.⁹ The demyelination occurs secondary to the ischemia but is also secondary to mechanical factors. The pain impulses associated with the local demyelination are thought to be ectopic foci of spontaneous activity that in turn is associated with ephaptic (fiber-to-fiber) conduction.¹⁰ With continued marked compression, Wallerian degeneration of the nerve occurs. There is persistent interference of intraneural microcirculation, with permanent epineurial and intrafascicular edema. The edema leads to increased endoneurial fluid pressure, and the epineurial edema forms a base for invasion by fibroblasts. A constricting epineurial scar may develop. There is generally degeneration, particularly of large fibers, whereas others present with local conduction block due to local demyelination. Recovery from severe injury is poor, and recovery probably represents those fibers with conduction block only.

Pudendal neuralgia impacts quality of life. Many patients are disabled and unable to perform the usual tasks of daily life, particularly if sitting is involved. In our modern society, many have office jobs and travel can be frequent and long.

Unfortunately, therapeutic outcomes are less than desirable. In this limited study, only 31% received benefit from pudendal nerve injection. Labat et al¹¹ reported on therapeutic blocks of the pudendal nerve. Thirty-nine patients had 1 month or less benefit, 6 had 3 months, and 1 for more than 3 months.¹¹

Sato and Nagai¹² reported sacral magnetic stimulation in 5 patients with pudendal neuralgia who had a single session of 30-50 pulsed stimuli. Pain was eliminated and relief lasted between 30 minutes and 56 days with a median of 24 hours.

The literature on carpal tunnel release supports surgical intervention in patients with evidence of axonal loss. In carpal tunnel a combination of clinical and electrodiagnostic findings rather than either alone should be used. Prolonged latencies with reasonably preserved amplitudes or conduction block in the carpal tunnel suggest local demyelination and potentially rapid recovery after surgery. Severity of symptoms does not correlate with latencies. Signs of axonal loss (sensory loss, muscle weakness and wasting on clinical exam with decreased amplitudes, fibrillation potentials, decreased recruitment, and increased size motor units) are signs of more severe compression. Patients with normal findings, in whom false-negative results can occur, have very mild compression, if any, and if clinical features are not typical, benefits of surgery should be carefully consid-

ered.¹³ This evidence can probably be extrapolated to patients with pudendal nerve decompression.

In 1994 in France, the group of Robert et al¹⁴ modified their surgical technique to a posterior approach. A perineal route in 40 patients with 48 operated nerves with 6-month to 7-year follow-up revealed improvement in 67% and no change in 33%. There was good relief in 16%, some in 58%, and none in 30%. In 1998 they reported that 70% of 170 operated patients had "good" results.¹⁵ Mauillon et al¹⁶ decompressed the pudendal nerve between the sacrotuberous and sacrospinous ligaments. Of 12 patients, 3 were cured, and 1 was slightly improved. The rest had no improvement. The 3 who were cured had at least 2 weeks of pain disappearance after nerve blocks prior to surgery, repeated twice.

This study was limited by retrospective review of charts, subjective reporting of pain, lack of randomization, and small numbers in the therapy groups.

In this retrospective, descriptive review, we reported the major clinical and neurophysiologic findings in patients with pudendal neuralgia. Furthermore, we described the outcomes of the medical and surgical management. Although most patients receive some benefit from conservative therapies, many patients seek a correct diagnosis and treatment, which will allow them to lead normal lives. This study revealed that 83% of these patients were misdiagnosed.

Pudendal neuromodulation was introduced as a possible therapy but requires further evaluation. A prospective study at centers with capabilities for electrodiagnosis and experience in medical and surgical management is needed to determine the best therapy for this severe pain syndrome.

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