Authors:

Christopher Edward Ramsden, BS
Michael Craig McDaniel, MD
Robert L. Harmon, MD
Kenneth M. Renney, MD
Alexis Faure, MD

Affiliations:

From the School of Medicine (CER, MCM) and the Departments of Medicine and Neurology (RLH), Medical College of Georgia, Augusta, Georgia; the Department of Orthopedics and Sports Medicine Associates, Houston, Texas (KMR); and the Department of Neurotraumatologie, Centre Hospitalier Universitaire, Nantes, France (AF).

Correspondence:

All correspondence and requests for reprints should be addressed to Christopher E. Ramsden, BS, 8250 Nesbit Ferry Road, Atlanta, GA 30350.

Pain

Review & Case Report

Pudendal Nerve Entrapment as Source of Intractable Perineal Pain

ABSTRACT

Ramsden CE, McDaniel MC, Harmon RL, Renney KM, Faure A: Pudendal nerve entrapment as source of intractable perineal pain. *Am J Phys Med Rehabil* 2003;82:479–484.

Perineal pain caused by pudendal nerve entrapment is a rarely reported entity, with only a handful of cases in the modern literature. A 25-yr-old male medical student had refractory unilateral orchialgia for 32 mo and concomitant proctalgia for 14 mo. Pain was positional in nature, exacerbated by sitting and partially relieved when standing or recumbent. Pudendal nerve entrapment was diagnosed clinically, with computed tomography–guided nerve blocks providing temporary relief. A prolonged left pudendal nerve distal motor latency on electrodiagnostic testing later confirmed the diagnosis. At surgery, the left pudendal nerve was found flattened in the pudendal canal of Alcock and in contact with the sharp inferior border of the sacrospinous ligament. After surgical decompression and rehabilitation, the patient experienced significant relief of pain and returned to medical school. This case suggests pudendal nerve entrapment should be considered in the differential diagnosis of chronic urogenital or anorectal pain, particularly if the pain is aggravated by sitting or if there is a history of bicycle riding.

Key Words: Pudendal, Neuralgia, Orchialgia, Proctalgia, Levator Ani Syndrome, Prostatodynia, Pain, Pelvic, Perineal

Although idiopathic urogenital and anorectal pain syndromes are not uncommon, effective treatments remain elusive for this patient group. Pain in these areas of the body can be embarrassing for the patient, limiting the desire to discuss the symptoms with the physician; physicians also may not be familiar enough with these pain syndromes, leading to misdiagnosis.[1] Poorly understood pain syndromes, including idiopathic vulvodynia, prostatodynia,

idiopathic orchialgia, idiopathic proctalgia, coccydynia, levator ani syndrome, and urethral syndrome, share many common features, including pain in the distribution of one or both pudendal nerves. A case report is presented to suggest that pudendal nerve entrapment (PNE) can be a cause of chronic perineal pain refractory to conventional treatments. The relevant literature is also reviewed.

CASE DESCRIPTION

A 25-yr-old male medical student developed severe, intractable, and progressive left-sided orchialgia. The pain was exacerbated in the seated position. Pain decreased when standing, recumbent, or when seated on a lavatory seat. The patient knew of no definite inciting event but believed pain onset may have resulted from several cycling workouts on a stationary bike. He denied any low back pain, focal weakness, or other associated illness. After about 18 mo, the patient began experiencing proctalgia and dyschezia; standing no longer provided relief. At 20 mo, a blind steroid/local anesthetic injection was performed near the left pudendal canal of Alcock via a perineal approach. The pain disappeared for about 2 hr but then returned. Pertinent medical history included right-sided "sciatica" 7 yr previously, associated with an L5–S1 disk herniation, with complete symptom resolution after hemilaminectomy and diskectomy.

Over a 32-mo period, this patient saw 15 different healthcare professionals (a family practitioner, emergency medicine physician, three urologists, two neurologists, two physiatrists, two pain specialists, an orthopedic surgeon, two physical therapists, and a chiropractor), had ten imaging tests performed, and was given six different diagnoses, including idiopathic orchialgia, nonbacterial chronic prostatitis, prostatodynia, levator ani syndrome, idiopathic proctalgia, and pudendal neuralgia (Table 1). Physical examination was unremarkable except for tight pelvic side wall musculature and increased external anal sphincter tone on digital rectal exam. Digital rectal exam with pressure on the left ischial spine reproduced unilateral perineal and rectal pain, presumably secondary to nerve compression of the pudendal nerve trunk. The patient stated that his present pain was a 9 on a 10-point visual analog pain intensity scale and was consistently >6. Between the 28th and 29th months, four guided (three using computed tomography [CT], one under fluoroscopy) steroid/local anesthetic injections were performed, two at the level of the ischial spine and two at the level of the pudendal canal (Fig. 1[2–4]; all four provided short-term relief but failed to provide long-lasting benefit. By 31 mo, the patient's pain was so severe that he was bedridden.

Initial electromyographic study of the ischiocavernosus muscle at 27 mo showed no evidence of denervation involving this muscle. At 32 mo, a left pudendal nerve distal motor latency was obtained.(Fig. 2).[2–4] The normal value for the perineal branch used for the laboratory was <4.0 msec; >5.0 msec was considered pathologic.[2,3] Pudendal nerve motor conduction studies utilized a St. Mark's pudendal electrode (London, UK) that normally incorporates stimulating and recording electrodes on


TABLE 1

Studies, preliminary diagnoses, and ineffective management approaches in the case presented to illustrate the initial clinical confusion in assessing the patient's symptoms

Negative Laboratory Tests	Unremarkable Diagnostic Imaging	Preliminary Diagnoses	Unnecessary Surgery	Failed Nonsurgical Treatments
1) ESR	1) Testicular ultrasound	1) Idiopathic orchialgia	Laparoscopic varicocelectomy	1) Lumbar traction
2) ANA	2) Prostatic ultrasound	2) Nonbacterial prostatitis		2) Myofascial release
3) B12	3) MRI lumbar spine	3) Levator ani syndrome		3) Biofeedback
4) Folate	4) MRI pelvis	4) Idiopathic proctalgia		4) Pelvic/lower extremity stretching
5) TSH	5) Abdominal and pelvic CT	5) Pudendal neuralgia		
6) CBC	6) Pelvic X-ray			
7) VDRL	7) Intravenous pyelogram			
8) Urinalysis with culture	8) Voiding cystourethrogram			
9) Semen analysis for leukocytospermia	9) Barium enema			

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Figure 1: Computed tomography–guided bilateral pudendal nerve block at level of the ischial spine. A 22-gauge needle is advanced through the gluteus maximus medial to the ischial spine to deliver local anesthetics and corticosteroids as described by Dr. Bensignor. Here, the nerve may be compressed by the sacrotuberous ligament, sacrospinous ligament, or both. The block has both diagnostic and therapeutic utility. Reproduced with consent from Dr. M. Bensignor.[2–4]


0.2 mV
5 ms

Lat. = 6.0
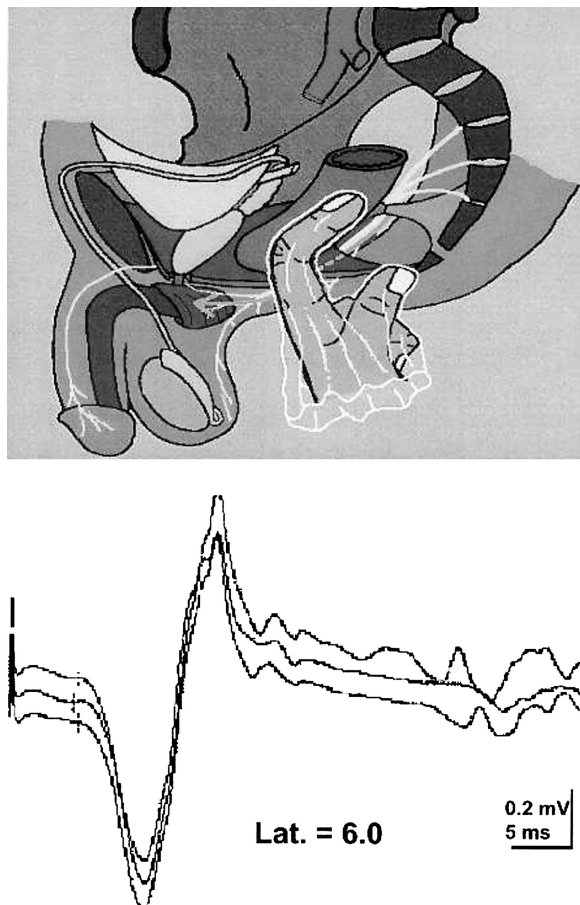
Figure 2: Pudendal nerve distal motor latency technique used in the case presented. The pudendal nerve distal motor latency was obtained via intrarectal stimulation using a St. Mark's Hospital Electrode (London, UK) applied at the left ischial spine and recorded through a needle electrode in the left bulbospongiosus muscle. Reproduced with consent from Dr. J. J. Labat.[2–4]

an adhesive sheet that fits on a gloved index finger that is then placed intrarectally.[5] In the case presented, however, a needle recording electrode was used in the left bulbospongiosus muscle. Only latencies were determined given amplitude variation from changes in recording electrode placement. Left pudendal nerve distal motor latency was 6.0 msec, supporting a diagnosis of left pudendal neuropathy; the right pudendal nerve was not studied at that time. Of note, earlier nerve conduction studies before the pudendal nerve injections might have been more valuable because the injections may have damaged the nerve.

The patient was then admitted for unilateral surgical exploration and decompression of the left pudendal nerve. With the patient in a ventral decubitus position, a vertical 8-cm incision was made in the left buttock lateral to the sacrum, exposing the gluteus maximus muscle fibers. These fibers were separated, and the sacrotuberous ligament was windowed to reveal the pudendal neurovascular bundle. The pudendal nerve trunk was flattened throughout most of the pudendal canal and compressed by the sharp inferior border of the sacrospinous ligament, straddling the ligament at an acute angle. The neurovascular bundle was moved medially, and the lateral aspect of the sacrospinous ligament was divided, just medial to the ischial spine. The thick obturator internus fascia forming the pudendal canal of Alcock was then divided, and the flattened nerve trunk was transposed anterior to the ischial spine.

After surgical decompression, the patient gradually improved. By 11 mo postsurgery, he was able to return to medical school. Although the pain was still occasionally exacerbated in the seated position, it was much less intense and subsided more rapidly after standing. At 19 mo postsurgery, pain averaged <1 on a 10-point visual analog pain intensity scale with-
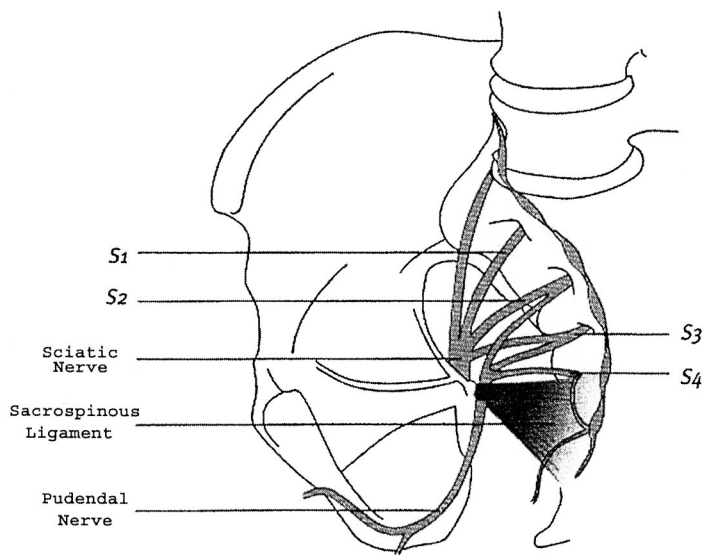
Figure 3: Anatomy of the pudendal nerve. This figure shows the pudendal nerve exiting the pelvis via the greater sciatic foramen and re-entering immediately adjacent to the inferior border of the sacrospinous ligament. Pudendal nerve entrapment can occur between the sacrospinous and sacrotuberous ligaments or in Alcock's canal. Reproduced with consent from Dr. J. J. Labat.[2–4]

out medications, and the patient was able to exercise regularly. (Of note, the patient underwent right pudendal nerve decompression surgery 13 mo after the left surgery for less severe pain. An anatomic anomaly of right pudendal nerve trunk was observed. The flattened trunk penetrated the sacrospinous ligament on its course to the perineum. Complete relief of right-sided symptoms was achieved immediately after surgery.)

DISCUSSION

The pudendal nerve arises from the ventral primary rami of S2, S3, and S4 of the sacral plexus (Fig. 3.[2–4] The pudendal nerve exits the pelvic cavity under the piriformis muscle through the greater sciatic foramen and descends ventral to the sacrotuberous ligament. The nerve then passes under the sacrospinous ligament medial to the ischial spine and re-enters the pelvic cavity through the lesser sciatic foramen.[2] While under the levator ani muscle, the pudendal nerve courses ventrally

through the pudendal canal of Alcock, a thickening of the obturator internus fascia. Within the ischioanal fossa, the pudendal nerve gives off two branches: the inferior rectal branch and perineal branch. The dorsal sensory nerve of the penis or clitoris forms the terminal branch.[2,6] The inferior rectal branch of the pudendal nerve passes medially through the ischioanal fossa, providing motor supply to the external anal sphincter and sensory supply to perianal skin. The perineal branch exits from the distal portion of the pudendal canal before splitting into the superficial perineal and deep perineal nerves.[2] The superficial perineal branch supplies sensation to the perineum and posterior aspect of scrotum or labia. The deep perineal branch of the perineal nerve provides motor supply to the bulbocavernosus, ischiocavernosus, superficial and deep transverse perineus, and sphincter urethrae muscles. The dorsal nerve of the penis or clitoris, the terminal branch of the pudendal nerve, courses ventrally under the symphysis pubis before

providing sensation to penile or clitoral skin excepting the dorsal surface at the base (which is innervated by the ilioinguinal nerve).[7] The two documented sites of PNE are between the sacrotuberous/sacrospinous ligaments and in the pudendal canal.[2,3,8]

Pudendal neuropathy as a source of fecal incontinence and occasional constipation and temporary loss of penile sensation, and even impotence has been described.[9,10] There is also limited literature identifying pudendal neuropathy as a possible source of perineal pain. Cases of pudendal neuralgia caused by solitary neurofibroma[11] and postherpetic pudendal neuralgia[12] have been described. In 1993, Hagen[13] presented six patients with diabetes, cancer, or both who had pain in the rectum or genitals attributed to pudendal neuralgia. In 1996, Alevizon and Finan[14] described one case of PNE after sacrospinous colpopexy; 2 yr after surgery, removal of a suture from the sacrospinous ligament produced total pain relief. In 2001, Amarenco et al.[15] described six patients with pudendal neuralgia secondary to various surgical procedures involving surgical traction. In addition, formerly idiopathic cases of vulvodynia later attributed to pudendal neuralgia have been presented.[16–18]

Amarenco et al.,[8,19,20] in 1987 and again in 1989 and 1997, identified entrapment neuropathy of the pudendal nerves as cause of chronic perineal pain. The syndrome was characterized by pain in the distribution of the pudendal nerves, particularly in the seated position, in absence of sexual disturbances. In men, the pain can affect the perineum, scrotum, anus, and penis. The pain can be unilateral or bilateral; pain intensifies during sitting and is diminished when standing, recumbent, or sitting on a lavatory seat. A relationship is recognized between burning pain and prolonged mechanical compression of the pudendal nerve within the pudendal canal shortly before symptoms begin.[21] Bicyclists,

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both professional and amateur, seem to be at increased risk of developing the syndrome, presumably due to chronic perineal microtrauma and resulting inflammation or fibrosis in the pudendal canal and the sacrotuberous/sacrospinous ligaments.

In 1998, Robert et al.[2] described in detail the mechanisms and consequences of entrapment of the pudendal nerves. After cadaveric studies in the seated position and >150 surgical explorations and decompressions (begun in 1987), two principal sites of nerve entrapment were identified: between the sacrotuberous and sacrospinous ligament and in the pudendal canal. Some nerves were found to be in conflict at both sites. Through a transgluteal surgical approach, surgical release achieved 67% success (45% cures and 22% significantly improved); factors that increased chances of success were shorter duration of pain, <50 yr of age at surgery, and a damaged nerve found on exploration. Factors with decreased success were >70 yr of age, pudendal nerve distal motor latency of >7.0 msec, or a normal nerve found on exploration. The unimproved 33% were not adversely affected.[2–4]

In 1998, Shafik[22] described 11 patients with idiopathic vulvodynia on whom pudendal canal decompression was performed via a perineal route, leaving the sacrotuberous and sacrospinous ligaments intact; 81% noticed significant improvement, with pudendal nerve distal motor latency reportedly normalized in 81%. In a 1999 article, Mauillon et al.[23] examined surgical outcomes of 12 patients with various chronic urogenital and rectal pain syndromes. These twelve patients were operated on via a gluteal approach, decompressing the sacrotuberous and sacrospinous ligaments and transposing the nerves proximal to the ischial spine. Of note, the pudendal canal was left intact. Three of the 12 patients (25%) were cured, and one was partially relieved at 21 mo.

Guided corticosteroid or local anesthetic blocks of the pudendal nerve have also been used for diagnosis and treatment of various perineal pain syndromes. In 1999, Thoumas et al.[24] described a CT-guided technique for blocking the pudendal nerve at potential entrapment sites. With the patient prone, a 22-guage needle is advanced through the gluteus maximus to either the ischial spine or the pudendal canal to deliver local anesthetic and corticoids. In 2000, Bensignor et al.[4] described a fluoroscopic or CT-guided pudendal nerve block technique at the level of the ischial spine and a CT-guided pudendal canal technique. Description of the pudendal canal technique emphasizes the necessity of angulation of the needle, which must be parallel to the plane of the obturator internus fascia. The average patient received 2.2 infiltrations. Reportedly, >65% of patients retained relief 1 yr after the last infiltration.[2,4] McDonald and Spigos[25] and Calvillo et al.[26] have described therapeutic and diagnostic utility of CT-guided nerve blocks for vulvodynia and anoperineal pain, respectively.

This case report illustrates the difficulty patients and physicians may encounter relating to various urogenital and anorectal pain syndromes. Physical exam did not provide many clues to etiology, despite the severe nature of the patient's pain. Reproduction of pain with digital rectal exam and pressure at the level of the left ischial spine was the only distinctive feature of the physical exam. The history, including the positional nature of the pain and relief when sitting on a lavatory seat, were the most important diagnostic clues.[2,8,19] The diagnosis was validated via CT-guided nerve blocks at the level of the ischial spine and pudendal canal. Nerve conduction studies, using an intrarectal technique to measure the distal motor latency, objectively confirmed the diagnosis. On surgical

exploration, the left pudendal nerve was found to be entrapped and flattened, with subsequent symptomatic improvement after decompression suggesting that PNE was the source of the patient's urogenital and anorectal pain.

CONCLUSION

PNE can cause pain in the rectum, scrotum (labia), or penis (clitoris). At onset, pain is exacerbated in the seated position, except on a lavatory seat, and relieved by standing or lying down. Diagnostic techniques, including CT-guided nerve blocks and nerve conduction studies, can confirm the diagnosis. Once diagnosed, guided corticosteroid injections and surgical intervention can provide relief and even cures for this formerly refractive population. At present, it is unknown what fraction of cases of idiopathic urogenital and anorectal pain syndromes are related to PNE. However, because accurate diagnostic methods and effective treatment options for PNE do exist, these diagnostic methods may be appropriate before concluding a patient has an idiopathic perineal pain syndrome.

ACKNOWLEDGMENTS

REFERENCES

1. Wesselmann U, Burnett AL, Heinburg LJ: The urogenital and rectal pain syndromes *Pain* 1997;73:269–94

2. Robert R, Prat-Pradal D, Labatt JJ, et al: Anatomic basis of chronic perineal pain: Role of the pudendal nerve. *Surg Radiol Anat* 1998;20:93–98

3. Robert R, Bensignor M, Labatt JJ, et al: Chronic perineal pain and the pudendal

nerve [in French]. *Ann Orthop Ouest* 1999;31:71–74

4. Bensignor M, Labat JJ, Robert R, et al: Perineal pain, anorectal, and urogenital [in French]. *Douleurs* 2000;1:131–142

5. Benson JT: *Pelvic Floor Neurophysiology: An AAEM Workshop*. Rochester, MN, American Association of Electrodiagnostic Medicine, 1998

6. Shafik A, Doss SH: Pudendal canal: Surgical anatomy and clinical implications. *Am Surg* 1999;65:176–80

7. Last RJ: *Anatomy: Regional and Applied*, ed 6. NewYork, Churchill Livingstone, 1978, pp 357–8

8. Amarenco G, Savatovsky I, Budet C, et al: Perineal neuralgia and Alcock's canal syndrome [in French]. *Annales de Urologie* 1989;23:488–92

9. Oberpenning F, Roth S, Leusmann DB, et al: The Alcock syndrome: Temporary penile insensitivity due to compression of the pudendal nerve within the Alcock canal. *J Urol* 1994;151:423–5

10. Anderson KV, Bovim G: Impotence and nerve entrapment in long-distance amateur cyclists. *Acta Neurol Scand* 1997;95:233–40

11. Tagnetti F, Poppi M, Gaist G, et al: Pudendal neuralgia due to solitary neurofibroma: Case report. *J Neurosurg* 1982;56:732–3

12. Howard EJ: Postherpetic pudendal neuralgia. *JAMA* 1985;253:2196

13. Hagen NA: Sharp, shooting pain in the rectum or genitals: Pudendal neuralgia. *J Pain Symptom Manage* 1993;8: 496–501

14. Alevizon SJ, Finan MA: Sacrospinous colpopexy: Management of postoperative pudendal nerve entrapment. *Obstet Gynecol* 1996;88:713–5

15. Amarenco G, Ismael SS, Bayle B, et al: Electrophysiologic analysis of pudendal neuropathy following traction. *Muscle Nerve* 2001;24:116–9

16. Turner ML, Marinoff SC: Pudendal neuralgia. *Am J Obstet Gynecol* 1991;165: 1233–6

17. McKay M: Vulvodynia: Diagnostic patterns. *Dermatol Clin* 1992;10:423–33

18. Jones KD, Lehr ST: Vulvodynia: Diagnostic techniques and treatment modalities. *Nurse Pract* 1994;19:37–46

19. Amarenco G, Kerdraon J, Bouju P: Treatments of perineal neuralgia caused by involvement of the pudendal nerve [in French]. *Revue Neurologique* 1997;153: 331–4

20. Amarenco G, Lanoe Y, Perrigot M, et al: A new canalar syndrome: Compression of the pudendal nerve in the pudendal canal of Alcock with perineal paralysis in cyclists [in French] *Presse Med* 1987;16: 399

21. Pisani R, Stubinski R, Datti R: Entrapment neuropathy of the internal pudendal nerve: Report of two cases. *Scand J Urol Nephrol* 1997;31:407–410

22. Shafik A: Pudendal canal syndrome as a cause of vulvodynia and its treatment by pudendal nerve decompression. *Eur J Obstet Gynecol Repro Biol* 1998;80:215–220

23. Mauillon J, Thoumas D, Leroi AM, et al: Results of pudendal nerve neurolysis: Transposition in twelve patients suffering from pudendal neuralgia. *Dis Colon Rectum* 1999;42:186–92

24. Thoumas D, Leroi AM, Mauillon J, et al: Pudendal neuralgia: CT-guided pudendal nerve block technique. *Abdom Imaging* 1999;24:309–312

25. McDonald JS, Spigos DG: Computed tomography-guided nerve block for treatment of pelvic pain due to pudendal neuropathy. *Obstet Gynecol* 2000;95:306–9

26. Calvillo O, Skaribas IM, Rockett C: Computed tomography-guided pudendal nerve block: A new diagnostic approach to long-term anoperineal pain. *Reg Anesth Pain Med* 2000;25:420–3

CME Self-Assessment Exam

Answers

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1. D

2. A

3. A

4. C

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