CHRONIC PELVIC PAIN: THE PERIPHERAL NEUROPATHIC BASIS

Stanley J. Antolak Jr, MD

Introduction and Disease Definition

Chronic pelvic pain (CPP) is a "boundary disease" affecting many specialties. It often has a neuropathic basis. The major neuropathy is a mononeuropathy of the pudendal nerve, a tunnel syndrome.[1] Pudendal neuropathy is a polysymptomatic phenomenon causing chronic, nonmalignant pelvic pain. It is one of the "great imitators" of modern medicine, which French experts call the "king of the pelvis." Pudendal neuropathy affects bowel, bladder, and sexual function, earning it the title of the "social nerve." Although a discussion of pudendal neuropathy (pudic nerve) was published in England in 1863,[2] and contemporary focus began in the 1980s in France and Egypt, diagnosis and treatment of pudendal neuropathy remain in a nascent status, remarkably ignored by most pain researchers. Most publications regarding pudendal neuropathy are case studies or cohort studies, with few controlled studies.

In its simplest form, pudendal neuropathy causes perineal pain (pudendal neuralgia) that is aggravated by sitting or driving, reduced when standing, and relieved by sitting on a toilet seat.[3] CPP is complicated by autonomic dysfunctions, central sensitization, and, occasionally, allostatic overload.

Symptoms of CPP confound specialists in gynecology, urology, physiatry, colorectal surgery, pain medicine, and neurology, as well as primary care physicians [*see Table 1*]. Using the taxonomy of the International Association for the Study of Pain (IASP), this review provides definite diagnoses for the neuropathic basis of CPP.[4] A definite diagnosis permits focused, appropriate treatments.

Chronic neuropathic pelvic pain is also caused by neuropathies affecting the anterior and posterior rami of thoracolumbar and sacral nerves [*see Table 2*].

Background

The impact of CPP is serious, equal to acute myocardial infarction, unstable angina, and acute ulcerative colitis.[5] CPP becomes a fatal disorder when patients commit suicide to achieve pain control. End-organ specialists who treat the suspected causal organs or tissues are generally the initial patient contacts. Despite repetitious complaints of pain and organ dysfunction, the diagnosis of neuropathic pain is delayed for years because evaluations and treatments are focused on suspected morphologic causes.

Definitions of CPP among specialty organizations treating CPP have similarities:

- The American Board of Obstetrics and Gynecology (ABOG) includes the following in female CPPS: chronic noncyclical pain, of a duration of 6 months or more, in the pelvis, abdomen, lower back, or buttocks.[6]
- The National Institutes of Health describes in males "lower genitourinary symptoms, particularly pain in the perineum or genitalia, voiding symptoms, such as dysuria or frequency, and sexual dysfunction."[7]
- The European Association of Urology (EAU) includes "non-malignant pelvic pain perceived in structures of the pelvis in either men or women. It is often associated with negative cognitive, behavioral, sexual and emotional consequences as well as symptoms suggestive of lower urinary tract, sexual, bowel, pelvic floor or gynecological dysfunction."[8] Pain must be continuous or recurrent for at least 6 months. The EAU definition embraces 21 syndromes, foremost of which are pelvic pain syndrome, bladder pain syndrome, prostate pain syndrome, scrotal pain syndrome, and pelvic floor muscle pain syndrome.
- The International Continence Society defines CPPS as "the occurrence of persistent or recurrent episodic pelvic pain associated with symptoms suggestive of lower urinary tract, sexual, or, bowel or gynecological dysfunction. There is no proven infection or other pathology."[9]

The ABOG and EAU syndromes include pains at nonpelvic sites. Fourteen EAU CPP syndromes include "symptoms suggestive of lower urinary tract, sexual, bowel, pelvic floor or gynecological dysfunction."[8] Patient symptoms often meet the criteria for more than one syndrome.[8] Among patients with pudendal neuropathy, 64% have additional symptomatic peripheral neuropathies (see below). These have a common sacral cord origin or overlap into the pudendal territory [*see Table 2*]. These "perimeter" neuropathies are significant pain generators, stimulate somatovisceral reflexes, and perpetuate windup.

Most CPP sufferers have undergone multiple specialist referrals followed by repeated, duplicated imaging studies, blood tests, and diagnostic "oscopies" without identifying a morphologic or infectious basis of symptoms. Well-intentioned interventions have not controlled their pains. Worse, multiple surgeries are often performed, to no avail. When clinicians cannot find objective results, patients are frequently told that the pain "is in [their] head," that is, somatization or a functional pain syndrome. Those misnomers ignore the neuropathic causes of CPP.

Few randomized controlled studies are found regarding pudendal neuropathy. This review provides an evidence-based diagnosis of specific neuropathies achieved using focused questions, specific examinations, and objective measurement of neuropathy while monitoring treatment progress with validated symptom scores.

Epidemiology of Pudendal Neuropathy

Few clinicians or researchers examine specifically for pudendal neuropathy, so the incidence and prevalence are uncertain. Prevalence is measured at 1% in a general population to 20% in a gynecologic practice.[9,10] CPP is common, affecting 15 to 16% of

Scientific American Pain Management
DOI 10.2310/7900.15034
01/17

Table 1 Neuropathic Basis of CPPS: Role of Pudendal Neuropathy and Multiple Additional Peripheral Neuropathies

1. CPPS is defined differently by medical/surgical specialties. CPPS includes pain in the pelvic region, lower abdomen, lower back, and buttocks. In addition, CPPS is associated with bowel, bladder, and sexual dysfunction.
2. Pudendal neuropathy is the major neuropathic cause of CPPS. Several other peripheral mononeuropathies may cause chronic pelvic pain.
3. Variable damage to autonomic and somatic sensorimotor nerve fibers results in a broad spectrum of symptoms.
4. Office diagnosis of pudendal neuropathy is readily made using pinprick sensation in the pudendal territory and observation of the skin for sympathetic changes. Additional "perimeter" neuropathies are diagnosed by physical examination.
5. Neurophysiologic tests can confirm pudendal neuropathy. Tests include the pudendal nerve terminal motor latency test, the warm detection threshold test, somatic sensory evoked potentials, and electromyography.
6. Central sensitization is usually present in patients with pudendal neuropathy.
7. Pudendal neuropathy will respond to prevention of nerve damage, a series of three pudendal nerve perineural injections of corticosteroids and bupivacaine, and, if necessary, decompression surgery.
8. The multiple "perimeter" neuropathies found in 64% of pudendal neuropathy sufferers usually respond to combinations of anesthetic nerve blockade and postural correction exercises. Surgical interventions may be necessary.

CPPS = chronic pelvic pain syndrome.

Table 2 Multiple Peripheral Neuropathies that Affect Pelvic Pain*

Neuropathy	*Males*	*Females*
Posterior ramus		
Maigne syndrome (TLJ)	11.6%	57.6%
Middle cluneal neuropathy	35.3%	56%
T12 posterior cutaneous perforating branch	Frequent†	Frequent†
T12 posterior ramus	Frequent†	Frequent†
Anterior ramus		
Abdominal cutaneous nerve entrapment	5.8%	15.4%
Ilioinguinal-iliohypogastric unilateral	35.3%	11.5%
Ilioinguinal-iliohypogastric bilateral	23.5%	38.4%
Perineal branch of posterior femoral cutaneous nerve	Occasional†	Occasional†
Posterior femoral cutaneous nerve	Occasional†	Occasional†
Genitofemoral nerve	Infrequent†	Infrequent†

TLJ = thoracolumbar junction.
*Multiple peripheral neuropathies are pelvic pain generators and are found in 64% of patients with pudendal neuropathy (n = 50).
†Imprecise term indicating inconsistent recording of findings in medical records.

females.[11] It accounts for 8% of urologic office evaluations. The prevalence of "chronic prostatitis" (which may reflect neuropathic pelvic pain) has an incidence of 3%, with an incidence of 3.3 per 1,00 patient-years.[12] The age at symptom onset ranges from a 10-year-old boy to a 94-year-old female in our experience.

Etiology

Pudendal neuropathy is a cumulative trauma syndrome due to compression, stretch, and direct trauma.[13] It is typically an acquired phenomenon secondary to repetitive microtrauma. Seven international pudendal clinicians identified their five most frequent, proximate causes. The etiology is not limited to these processes; for instance, radiation neuropathy of the pudendal nerve is infrequent but devastating [*see Table 3*]. In the 21st century, the use of mesh during incontinence surgery has become the most common cause.

Psychiatric illnesses do not cause CPP; however, psychosocial disorders are common among sufferers. Secondary depression and anxiety occur because of the remarkable disabling nature of chronic pain. In many patients, emotional dysfunction follows several years of misdiagnosis, innumerable and repetitive tests, ineffective interventions, and a disrupted lifestyle. Disruption of the social network often requires psychological or psychiatric consultation. Integrated, comprehensive care might include relaxation, life coaching, and other techniques.

The central origin of CPP should be discounted. The hypothesis that abnormal processing of afferent sensory input causes symptoms such as pain or urinary urgency overlooks the peripheral neuropathic component of the pains of CPP sufferers.[14] Changes in the brain found by magnetic resonance imaging (MRI) or positron emission tomography (PET) more likely reflect the neurochemical effects of chronic noxious afferent stimuli.[15] Effective future research requires

Table 3 Causes of Pudendal Neuropathy

*Identified Cause**	*Incidents or Activities*
Cycling (7)	Bicycle seat; distance cycled; number of pauses
Prolonged sitting (7)	Computer tasks, seamstress, long automobile or airplane trips (truck drivers, salesmen), airplane pilots, attorneys
Pelvic surgery (7)	Gynecologic surgery (especially using mesh), radical prostatectomy. Any pelvic procedure may cause windup of previous pelvic pain.
Pelvic trauma (4)	Falls, motor vehicle accidents, motocross, horseback riding, sexual toys
Adult exercise (4)	Running, abdominal crunches, leg presses, weightlifting, spinning, triathlon, stair climbing, dance line, cheerleading, volleyball, track, soccer, football, hockey, baseball (catcher and second baseman), basketball
Traumatic childbirth (3)	Duration of second stage of labor; large-size infant
Constipation (2)	Usually prolonged over many years. Constipation may be the cause or result of pudendal neuropathy.

*Causes of pudendal neuropathy identified by seven experienced international "pudendal clinicians." Numbers in parentheses indicate the number of clinicians identifying their top five causes.

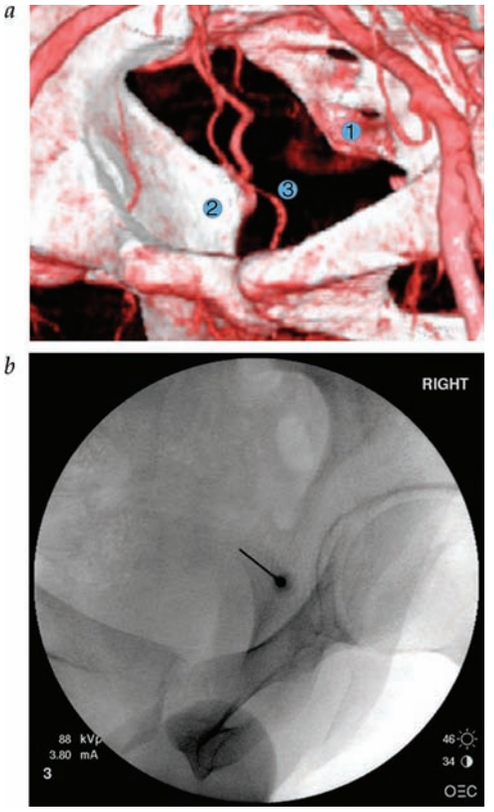

Figure 1 **(*a*) Common changes in bony remodeling in patients with pudendal neuropathy seen on a magnetic resonance arteriogram. 1 = Inferolateral angle projecting into the greater sciatic notch (GSN). 2 = Broad, flat tip of the remodeled ischial spine and ischium projecting into the GSN. 3 = Compression of the pudendal artery at the interligamentous space. (*b*) Fluoroscopic view of a pudendal nerve block demonstrating bony changes in the ischial spine: (1) the ischial spine, superior margin; (2) the femoral head; (3) the superior pubic ramus. The ischial spine is remodeled with a broad, sharp tip and "hooking" superiorly that forces the pudendal nerve superiorly and laterally. Surgery is quite difficult with this type of remodeling. Paresthesias during needle placement located the nerve at the inferior margin of the ischial spine.**

that a definite diagnosis of pudendal neuropathy be established in subjects and ruled out in controls.

Pathogenesis and Pathophysiology

Clinical symptoms suggest that pudendal neuropathy is mainly a disorder of small, unmyelinated C fibers affecting both somatic and autonomic afferent and efferent nerves. Nervi nervorum are also implicated in neuritic pain.

Causes of pudendal neuropathy include compression, stretch, direct trauma, and iatrogenic interventions such as pelvic surgeries (especially with mesh) and radiation [*see Table 3*]. A common theme among patients is repetitive flexion at the hip during running, cycling, exercising, and sports activities. Many patients were youthful athletes. Pressure on the perineum while sitting on a bicycle seat exceeds by twofold the pressure necessary to cause neural ischemia.[9]

The bipedal stature of humans demands a strong pelvic floor and a well-developed ischial spine versus quadrupeds or apes. Traction by ligaments and fascias remodel the ischium, the ischial spine, and the inferior lateral angle of the sacrum. The diameter of the greater sciatic notch is reduced.[16] Bony changes are consistently evident during fluoroscopic pudendal blocks, during Judet views of the pelvis, or with three-dimensional reconstruction of pelvic computed tomographic (CT) scans [*see Figure 1a*]. The remodeled ischial spine projects medially, superiorly, and posteriorly. Its tip may become broad and rounded or have very sharp edges [*see Figure 1b*]. Changes are usually bilateral. Unilateral remodeling occurs in equestrians and in athletes pushing off consistently with the same leg (e.g., football running back or pivoting in basketball). The attached sacrospinous ligament broadens and rotates. The interligamentary space narrows and restricts nerve movement or causes compression. Sitting compresses the nerve between the sacrotuberous and sacrospinous ligaments (the "lobster claw" or "clamp"), probably causing ischemia. In addition, the ischioanal fat body may elevate the nerve against the falciform process of the sacrotuberous ligament [*see Figure 2*].

Most surgical compressions (about 90%) are found between the sacrotuberous and sacrospinous ligaments (the interligamentous space). Multiple fascial bands may compress the nerve near the ischial spine. About 10% of compressions are observed in the pudendal (Alcock) canal. A hypertrophied piriformis muscle may force the nerve against the sacrospinous ligament. Two or more compression sites may be found. Nerve branches may penetrate the ligaments, commonly the inferior rectal nerve (IRN) in about 10% of cadavers/patients. The IRN exits prior to the Alcock canal in about 50% of patients,

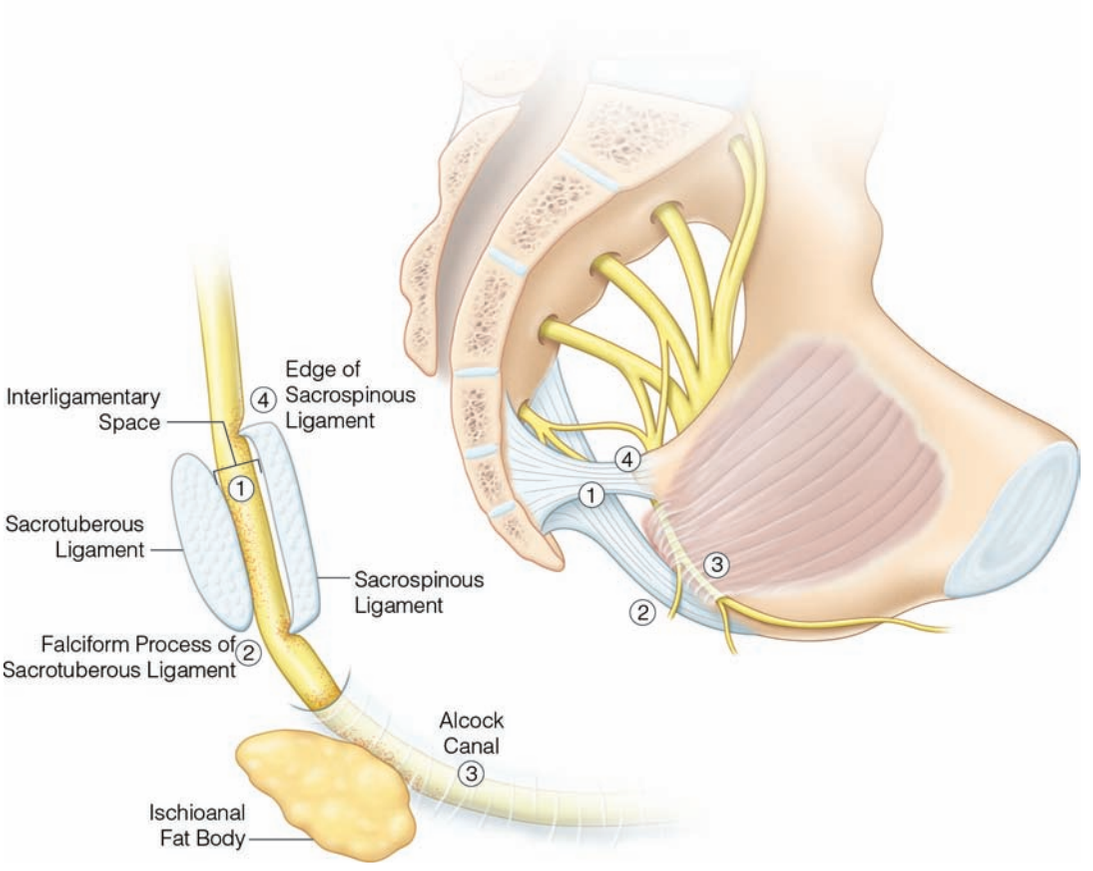

Figure 2 **Common sites of pudendal nerve compression. 1 = Compression at the interligamentary space. 2 = Compression against the falciform process by ischioanal fat body when sitting. 3 = Alcock canal. 4 = Conflict by the sharp edge of the sacrospinous ligament.**

preventing compression at that anatomic site. Nerve tethering by fascias leads to stretch when climbing stairs, during childbirth, and with chronic constipation.

At the pudendal canal, thickening of the obturator fascia may occur. When the obturator internus muscle hypertrophies, from exercise, it compresses the nerve medially into this fascia. Hypertrophy of the falciform process of the sacrotuberous ligament in athletes can cause impingement on one or more branches of the pudendal nerve.

Onset of pudendal symptoms concurrent with shingles infections is unusual. It is unclear whether symptoms are caused by a de novo infection of the nerve or perhaps represent windup of preexisting pudendal neuropathy. Patients treated for "prostatitis" often have pain reduction using quinolone antibiotics or minocycline. Less commonly, patients with definite pudendal neuropathy report testing positive for Lyme disease. They may report symptomatic improvement after antibacterial therapy for sinusitis, etc. The responses may be secondary to the analgesic effects of antibiotics, including tetracycline, minocycline, and quinolones.[17,18]

In our limited experience with paraplegic patients (n = 3), rectal pain due to prolonged sitting was considered sympathetically mediated pudendal neuropathic pain and responded to pudendal neural blockade. One patient required concurrent pelvic sympathetic neural blockade.

Anatomy

PUDENDAL NEUROANATOMY

Sacral cord levels S2, S3, and S4 generally provide somatic and parasympathetic innervation to the pelvis. The pudendal nerve originates from the sacral plexus. Fibers are chiefly somatic (70%), with approximately 50% sensory and 20% motor fibers. Autonomic fibers account for 30% of the nerve. The main trunk generally forms proximal to the ischial spine. It travels a serpentine route through the gluteal region and enters the pelvis medial to the ischial spine between the sacrotuberous and sacrospinous ligaments (the primary compression site). At the inferior margin of the ischial spine, the main trunk enters the pudendal or Alcock canal. This space, between duplicated layers of the fascia of the obturator internus muscle, is a second compression site. The nerve trifurcates as it continues distally to terminate in three branches: the dorsal nerve of the penis/clitoris, the perineal nerve, and the IRN [*see Figure 3*]. These provide sensory innervation to the genitals, perineum, and anococcygeal skin. At the level of the ischial spine, the IRN may branch from the main trunk (50%) prior to the Alcock canal [*see Figure 3a*]. The IRN is separate from the main trunk in 10% of patients, often piercing the sacrospinous ligament.

Complex variants in the origin of the pudendal nerve, pudendal plexus, and branches of the pudendal nerve account

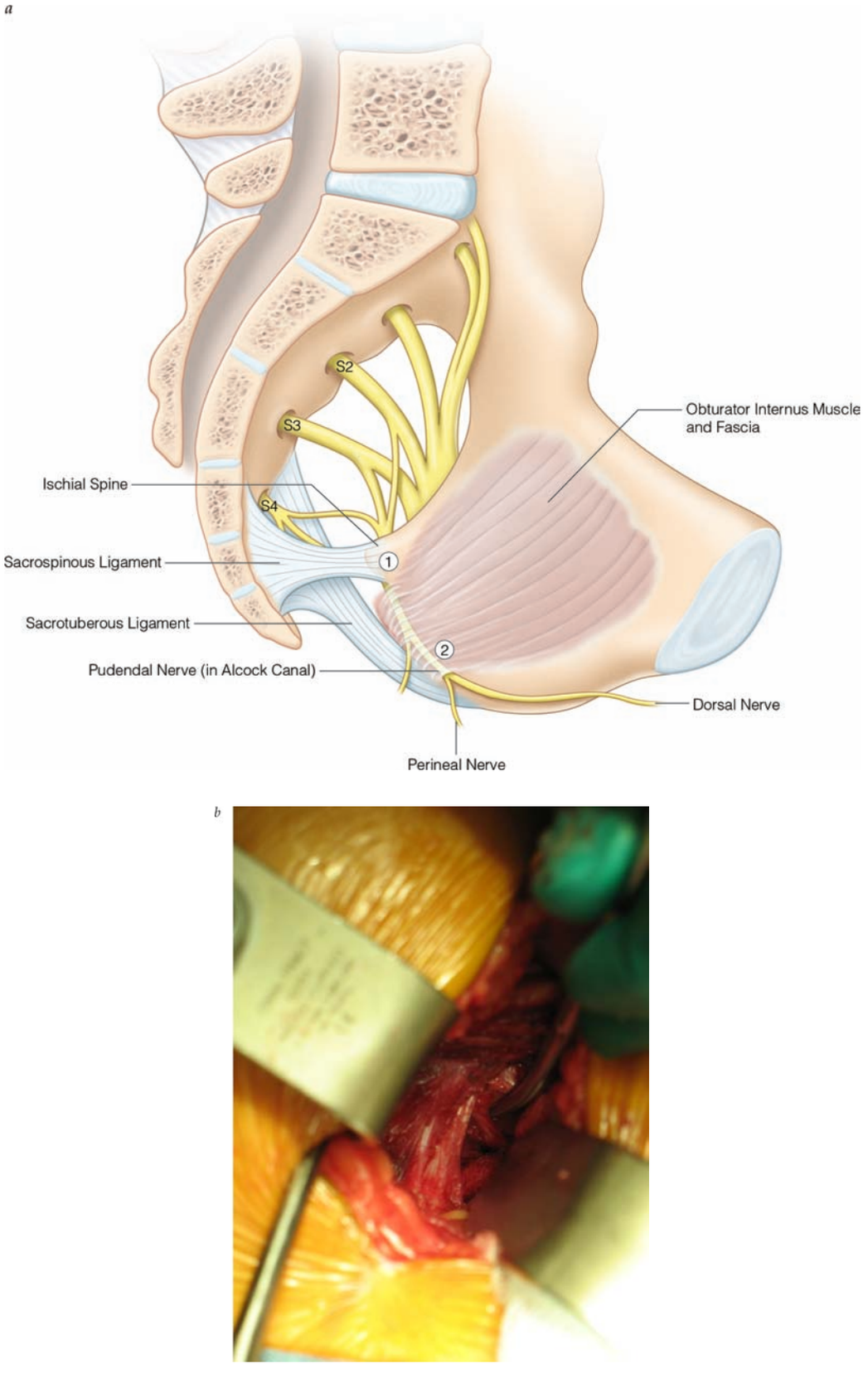

Figure 3 *(a)* **Anatomy of the pudendal nerve. After formation, the nerve trunk travels (1) posterior to the sacrospinous ligament medial to the ischial spine. At the inferior margin, it enters the Alcock canal (2) in the fascia of the obturator internus muscle.** *(b)* **Left falciform process. The left sacrotuberous ligament (opened longitudinally) sends unusually thick and firm fibers distally to form the falciform process that extends medially into the pelvis. It widens, covering the obturator internus muscle and the main trunk of the pudendal nerve. The inferior rectal nerve exits prior to the Alcock canal and is seen in the ischioanal fossa between the scissors and the retractor.**

for symptom disparities among patients and are reviewed below. During flares of pelvic pain, concurrent pain in the calves, feet, and toes may occur because the sciatic nerve may have fibers from the pudendal plexus in up to 26% of cadavers.

Autonomic dysfunction is a major component of pudendal neuropathy and accounts for many of the bowel, bladder, and sexual symptoms. Heart rate variability and increased blood pressure are consistently measured in both genders with CPP, including interstitial cystitis.[19,20] Pudendal somatic afferent fibers project onto sympathetic neurons in the thoracolumbar regions. Sympathetic efferent fibers travel via the inferior mesenteric ganglion and the presacral plexuses to the pelvic organs. Sympathetic aggravation of pelvic pain occurs with emotional stress, weather changes, humidity, or cold. Women sense an endocrine-mediated premenstrual aggravation of pain. An important clinical note is that pain cephalad to the vaginal introitus (i.e., dyspareunia with deep thrusting) and pain cephalad to the anal verge (i.e., rectal foreign-body sensation) are sympathetic, not somatic, in origin. Therapeutic importance includes the need for sympatholytic medications in many patients and, occasionally, injections of paravertebral or pelvic sympathetic ganglia. The ganglion impars has not been of significant clinical importance as measured by effectiveness following blocks.

Diagnosis

A clinical diagnosis is possible at the time of consultation in over 90% of patients and increases to 100% following neurophysiologic testing. [*See Figure 4*] Diagnosis is not made by laboratory blood tests or imaging. Most practitioners make a presumed diagnosis of pudendal neuropathy by analyzing patients' symptoms. Diagnosis is not made by a "check-off" list such as the Nantes criteria or by "diagnostic" pudendal blocks, as is discussed later.

CLINICAL MANIFESTATIONS

Compression of a mixed nerve may initiate a wide variety of somatic and autonomic symptoms. For instance, the classic pains of pudendal neuropathy are chiefly perineal and genital but may be suprapubic ("bladder pain"), sacral, coccygeal, and perianal.[3] Pain quality varies from burning pain to tearing, electric pulses, tugging, gnawing, and pressure. Paresthesias may occur. Allodynia is common. Dysuria, painful ejaculation, and painful bowel movements occur. Both genders may have sexual complaints. Pain may be initiated by sexual thoughts. Erectile dysfunction or anemission occur. Sexual sensation may be diminished or absent. Persistent genital arousal disorder occurs more often in females. Painful vaginal penetration may preclude intercourse. Symptoms may change over time; for example, after months or years, symptoms of an irritable bladder or retention of urine may require catheterization [*see Table 4*]. *Pudere* means to shame, and patients are often reticent regarding symptoms. Clinicians must ask multiple, direct questions to elicit all symptoms. Simple questions may suggest the possible nerve(s) involved in patients' pain [*see Table 5*].

A foreign-body sensation in the pelvis is frequent, such as sitting on an object (golf ball, nut), the presence of a red-hot poker in the vagina, broken glass in the rectum, or a red-hot bowling ball in the pelvis.

A number of factors affect pudendal pain location and variations between patients. These are discussed more thoroughly

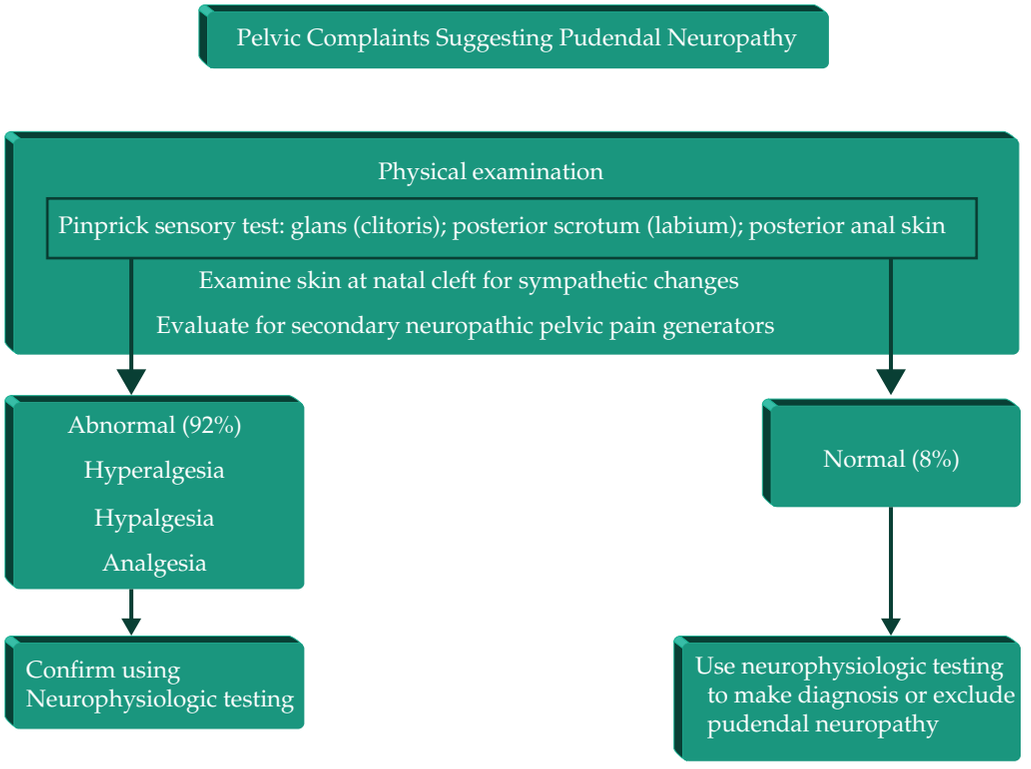

Figure 4 **Diagnostic algorithm for pudendal neuropathy.**

Table 4 Symptoms of Pudendal Neuropathy*

Pain	*Bladder*	*Bowel*	*Sexual*	*Central*
Perineal Clitoris Labia Vagina (introitus) Glans Scrotum Anal Suprapubic Coccyx Sacrum Inner thigh Motor Waddling gait Sphincter dysfunction	Urgency Frequency Incontinence Dysuria Persistent urge Hesitancy Slow stream Retention Pain with filling (sympathetic)	Diarrhea Urgency Frequent stools Incontinence Pain with urge Obstructed defecation (dyschezia)	Erectile dysfunction Painful ejaculation Anejaculation Persistent arousal in both genders Anorgasmia Painful orgasm Painful penetration Pain with deep thrusting (sympathetic)	Anorexia Nausea Diaphoresis Tremors Pains in calves and feet Tachycardia Hypertension

*A partial list of somatic and autonomic symptoms and allostatic overload defined by relief following treatment of pudendal neuropathy.

below. Receptive fields of multiple pelvic sensory nerves [*see Table 2*] overlap and may be painful.

Motor dysfunction includes a waddling gait due to impairment of the nerve to the obturator internus muscle, obstructed defecation, urinary retention, slow stream, or hesitancy.

Central sensitization has a great impact on symptoms. Pain in the legs, chiefly the toes, feet, and calves, occurs frequently. Symptoms get worse when pelvic pain is aggravated but gradually diminish and disappear as treatments proceed.

Allostatic overload is demonstrated in occasional patients with tachycardia, hypertension, profuse diaphoresis, and tremors. Gastroparesis and marked weight loss may occur. Disabling disruption of thought processes limits employment in some CPP sufferers, especially machinists, accountants, attorneys, and judges. Symptoms are controlled by treatments of pudendal neuropathy.

Parasympathetic dysfunction is not seen often. Patients must be asked about anal mucus secretion, excessive vaginal secretions, or any malodorous skin secretion.

PHYSICAL EXAMINATION FINDINGS

Pinprick sensory response to a safety pin is the premier office test for pudendal neuropathy. It tests C fibers and Aδ fibers. A safety pin is used. An abnormal response at any one of the six nerve branches indicates neuropathy.[21,22] Nerve branches are not damaged equally, and any patient may demonstrate mixed responses, including hyperalgesia, hypalgesia, analgesia, or normal [*see Table 6 and Figure 5*]. Patients are wary of pinprick in the genital region. It is essential to demonstrate the technique of light pinprick on the arm or thigh. Prior to examining each pudendal branch, a normal site is lightly touched. This repetitive technique, the continuous pinprick comparison method, permits the patient to have a consistent reminder of a normal response versus hyperalgesia, etc.[23]

Pinprick testing following therapeutic pudendal nerve blocks indicates the quality of the block. Responses may include analgesia, hypalgesia, or no anesthetic response at any of the six branches. An ideal block would provide analgesia at all six branches.[24]

Sympathetic dysfunction is readily recognized by vasomotor and sudomotor skin changes in the pudendal territory [*see Figure 6*]. The changes are typically centered over the natal cleft but can be seen on the perineum and at the transverse gluteal fold of the upper thigh/buttocks. Marked, evanescent "explosion" of skin changes may occur during pinprick examination, digital internal examination, or neurophysiologic testing. Changes normalize after a few seconds. Contraction of one or both labia is an important sign. The scrotum may resemble a tennis ball because of combined piloerector stimulation and contraction of the dartos muscle (sympathetic). Localized sweating is occasionally observed over the sacral area.

Internal Pelvic Examination

A digital rectal examination evaluates anal sphincter tone and tenderness, the prostate gland, the levator ani muscle, and the obturator internus muscles. Finger pressure medial to the ischial spine may reproduce subjective symptoms in about 30% of patients. This is the Valleix phenomenon, not a trigger point, and it indicates pudendal neuropathy. Prostate secretions or seminal fluid should be evaluated for the presence of inflammatory cells.

Females typically decline a digital rectal examination; however, the pelvic floor can be evaluated transvaginally along with the uterus and ovaries and the Valleix phenomenon. While in the gynecologic position, a pinprick sensory examination is performed and the skin is examined. The warm detection threshold

Table 5 Questions that Guide Practitioners to a Particular Neuropathy

Question	*Probable Nerve Involved*
Is perineal pain aggravated by sitting or driving?	Pudendal neuropathy
Is abdominal pain aggravated by waist band or belt?	Thoracolumbar junction syndrome, abdominal cutaneous neuropathy, iliohypogastric/ilioinguinal neuropathies
Is low back pain aggravated by bending forward to brush teeth or wash dishes?	Middle cluneal neuropathy
Do sexual thoughts cause or aggravate pain?	Central sensitization
Does wearing clothing cause pain?	Allodynia—a rapid indicator of neuropathy

Table 6 Female Pinprick Responses at Consultation*

Sensory Response	*Sites, n (%)*	*Right*			*Left*		
		Clitoris	*Labium*	*Anus*	*Clitoris*	*Labium*	*Anus*
Hyperalgesia	68 (43.5)	14	10	11	9	13	11
Normal†	39 (25)	3	9	8	6	6	7
Hypalgesia	28 (17.9)	7	4	2	9	4	2
Analgesia	21 (13.4)	2	3	5	2	3	6

*In 26 consecutive females, a total of 156 sites were tested with a safety pin (three bilaterally in each patient) and compared with normal pinprick sensation at the thigh.
†Two women had normal pinprick sensation at all six sites.

(WDT) test and the pudendal nerve terminal motor latency test (PNTMLT) are performed.

Additional Evaluations

Additional evaluations include observation of gait and pelvic tilt and a thorough physical examination for the additional "perimeter neuropathies" that affect CPP. These are discussed following the section on treatments of pudendal neuropathy [*see Table 7*].

LABORATORY TESTS

Imaging Studies and Blood Testing

Imaging studies cannot diagnose pudendal neuropathy. Normal MRI or computed tomographic (CT) scans will exclude other pathology. The physical examination is most important. A lack of abnormal findings in "routine testing" among CPP patients is not a reason to diagnose a functional somatic syndrome or somatization.

MRI evaluation is currently being evaluated as a means to identify changes in the pudendal nerve or perineural scarring. Findings have not been coordinated with surgical findings or with more conservative treatments. MRI needs to be studied after a definite diagnosis prior to recommending these expensive imaging tests routinely.

Blood tests for peripheral neuropathies are helpful only for exclusion.

The Nantes criteria attempt diagnosis by symptomatology but are contradicted by the evaluation findings outlined above.[26]

Neurophysiologic Testing for Pudendal Neuropathy

Neurophysiologic testing for pudendal neuropathy permits grading of patients' neuropathy from "possible" or "likely" to "definite."[4,26] The WDT test and PNTMLT are rapid and inex-

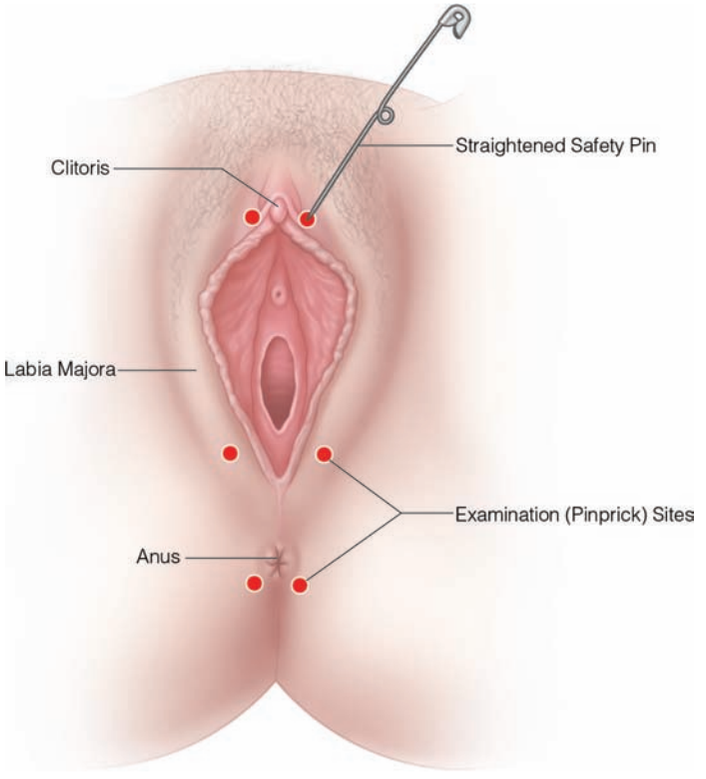

Figure 5 **Anatomic sites for sensory testing for pudendal neuropathy in females. Light touch with a safety pin at six examination sites measures all branches of the pudendal nerve.**

Table 7 Peripheral Neuropathies Frequently Identified in Patients with Pudendal Neuropathy*

Nerve	*Symptoms*	*Examination to Reproduce Pains*
Posterior rami of thoracolumbar junction (Maigne syndrome)	Abdominal pain; often suprapubic; elastic waistband is painful; bladder pain. May have flank pain and gluteal pain.	Skin rolling from flank to symphysis pubis and along buttock; often pressure on T12 vertebra causes local pain. Mild scoliosis is common.
Abdominal cutaneous	Abdominal pain	Pressure along rectus border bilaterally from T8 to T12
Ilioinguinal and iliohypogastric	Suprapubic and groin pain	Pressure at external inguinal ring above and below pubic tubercle
Middle cluneal	Sacral low back pain aggravated by forward bending, e.g., to brush teeth, lift a child	Pressure over S2, S3, S4 levels of sacrum medial to sacroiliac joint. In 30%, a palpable, movable lipoma (the "back mouse") can be palpated.
Perineal branch of posterior femoral cutaneous nerve	Gluteal, perineal, and groin pain	Pressure over posterior femur 4 cm below femoral head
T12 posterior ramus	Pain in hip, buttock, lower quadrant	Pressure over iliac crest 7.7 cm from midline
T12 posterior cutaneous perforating nerve	Pain at greater trochanter, buttock, lower quadrant, genitals	Pressure over iliac crest 8 cm posterior to anterosuperior iliac spine
Posterior femoral cutaneous nerve	Pain in posterior thigh	Pressure at superolateral margin of ischial tuberosity
Inferior gluteal nerve	Pain in lower buttock	Pressure at lower margin of gluteus maximus
Genitofemoral nerve	Pain in groin, genitals, lower quadrant	A diagnosis of exclusion
Obturator nerve	Pain in medial thigh	Pressure on nerve at digital pelvic examination

*Secondary peripheral neuropathic pain generators at the perimeter of the pudendal nerve receptive field require concurrent treatment when identified.

pensive and can be performed by any clinician or staffperson. In one recent review of our practice, all patients measured abnormal using these tests [*see Table 8*].

WDT Testing

WDT testing provides any clinician with a rapid and simple means for confirmation of pudendal neuropathy.[23] WDT testing evaluates cutaneous somatic sensation, providing quantitative evidence of small fiber neuropathy. "Qualitative" responses occur secondary to central sensitization. WDT testing correlates well with epidermal nerve fiber density.[27] The use of WDT testing for evaluation of pudendal neuropathy began after it was shown to be effective in identifying neuropathy of the dorsal nerve of the penis in impotent males.[28] WDT evaluation of all six branches of the pudendal nerve requires a small thermoprobe. WDT at consultation was abnormal at one or more branches in 92% of patients with symptoms suspected to be pudendal neuropathy.

The Physitemp NTE-2A (Physitemp, Clifton, NJ. USA) has a 1 cm diameter thermoprobe. small size correlates better with the number of epidermal nerve fibers in biopsy specimens.[29] Most commercially available probes are larger and may result in spurious results due to overlap into the territory of other nerves.[30] A stepping algorithm with increments of 4°C, 2°C, and1°C[31] begins at a neutral temperature, typically 31.5°C. Adjustment is needed if 31.5°C is warm or cool. Heat pain often occurs at 4°C increments, so our clinical testing is limited to 2°C and 1°C increments. To be conservative, the lower limit of abnormal sensation was selected as 39.5°C. This is 4 standard deviations from the reported normal of 37.4°C.[27] It approximates the upper limit of normal found by Beco and colleagues.[32]

Testing is performed in the dorsal lithotomy position. Males are tested at the glans (3 and 9 o'clock positions), posterior scrotum and posterior perianal skin, and anal verge [*see Figure 7*]. The dorsal nerve of the clitoris was tested at the base on each side or, when size necessitated, at the upper medial labia minora. WDT responses (unmyelinated C fibers) do not correlate with pinprick responses (Aδ). In general, detection of warmth is different at each nerve branch. Quantitative results of WDT testing vary from normal to more than 43.5°C, that is, no sensation of warmth at 43.5°C. Qualitative abnormalities indicate central sensitization and include dysesthesias (bladder warmth, rectal urge), paresthesias, and allodynia and occur at both normal and abnormal temperatures [*see Table 9*]. The limitations of quantitative sensory testing are discussed by Krumova and colleagues.[32] A supplement to WDT testing is to measure skin temperature with an infrared thermometer. Temperatures are typically lower in the neuritic territory, presumably due to abnormal sympathetic vasomotor stimulation.

Pudendal Nerve Terminal Motor Latency Testing

The PNTMLT has been used to assist diagnosis of pudendal neuropathy by colorectal and gastroenterology specialists and gynecologists, especially in patients with fecal incontinence.[34–36] PNTMLT identifies pudendal nerve function in women with refractory overactive bladder and nonobstructive urinary retention.[37] It measures only the motor component from the ischial spine distally. The St. Mark's surface electrode is placed medial to the tip of the ischial spine either transvaginally or transrectally. Multiple stimuli are recorded using Medtronic Sofomor Danek™ USA software. Changes in the latency, amplitude, and temporal dispersion are recorded.

Table 8 Neurophysiologic Testing*

Test	Right Side (%)	Left Side (%)	Abnormal Pinprick (%)
Males (n = 25)			
WDT	96	96	92
PNTMLT	88	92	—
Females (n = 26)			
WDT	53.8	61.5	92.3
PNTMLT	80.7	88.4	—

PNTMLT = pudendal nerve terminal motor latency test; WDT = warm detection threshold.
*Neurophysiologic testing results at consultation in patients with symptoms of pudendal neuropathy. Abnormalities are quantitative and/or qualitative. The WDT is abnormal at one or more sites. The PNTMLT is abnormal unilaterally or bilaterally.

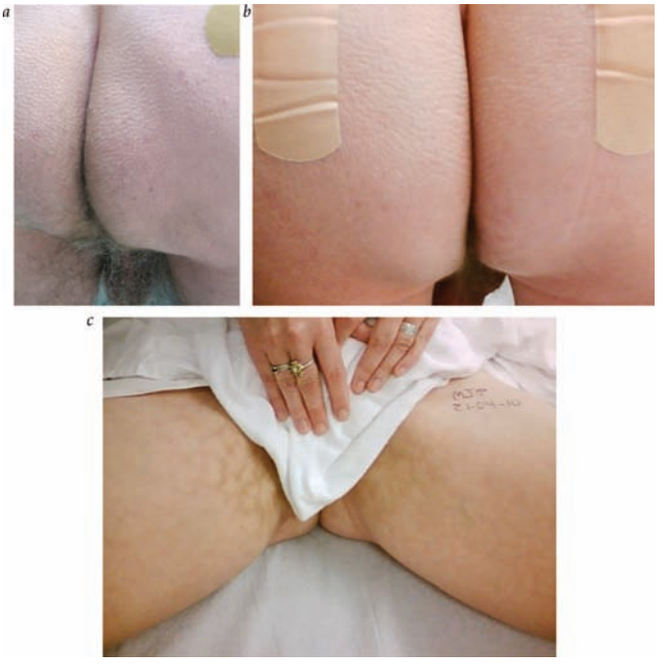

Figure 6 **Common vasomotor and sudomotor skin changes with pudendal neuropathy. Cutis anserina (*a*) is most common. Peau d'orange (*b*) is less common. Cutis reticularis (marmorata) (*c*) is infrequent. Sweating is not portrayed.**

Normal latency published by various authors ranges from 2.1 to 2.4 ms. PNTMLT can be performed by any practitioner. Slow conduction time, a "prolonged PNTMLT," demonstrates demyelination but is not specific for nerve compression. Intraoperative testing may show immediate improvement in the PNTMLT following decompression surgery [*see Figure 8*]. Shafik consistently demonstrated improvement in the PNTMLT postoperatively [*see Table 10*].[38]

Despite limitations, an abnormal PNTMLT confirms motor neuropathy between the ischial spine and the anus. PNTMLT is readily tolerated in both genders, but stimuli may cause significant pain in patients with central sensitization. A frequent site is the great toe of either foot.

Table 9 Warm Detection Threshold Results in a Female with Chronic Pelvic Pain*

Site tested	Right	Left
Clitoris	> 43.5°C	36.5°C
Labium	> 43.5°C; †felt pain in RLQ	41.2°C; †felt pain in LLQ
Anus	> 43.5°C; †felt pain in toes of left foot	37.4°C; †felt pain in toes of right foot

LLQ = left lower quadrant; RLQ = right lower quadrant.
*Abnormal quantitative and qualitative responses to warm detection threshold testing.
†Qualitative abnormalities occur at both normal and abnormal temperatures and indicate central sensitization. Note the contralateral response at normal temperature at the left inferior rectal nerve.

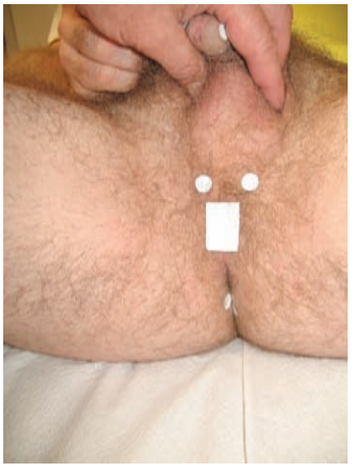

Figure 7 **Warm detection threshold test sites. The 1 cm diameter thermode placement avoids the midline because of overlapping fibers from the contralateral nerve. The large Medoc thermal probe crossing midline would measure both perineal nerves.**

Somatosensory Evoked Potentials

Somatosensory evoked potential (SSEP); testing is used more widely in Europe than in the United States.[39] Dr. Mark Conway (Nashua, NH; 2015) estimates that SSEP confirms neuropathy in approximately 80% of patients suspected of having pudendal neuropathy. The test also correlates similarly with identifiable compression of the pudendal nerve at decompression surgery.

Pudendal Electromyography

French neurophysiologists' experience with pudendal neuropathy dates from the 1980s. In the United States, Benson, a

Table 10 Postoperative Improvement in PNTMLT[37]

PNTMLT	Mean (ms)	Range (ms)
Before treatment	6.9 ± 1.3	5.1–7.6
After treatment	2.3 ± 0.3 (p < .05)	2.1–2.7

PNTMLT = pudendal nerve terminal motor latency test.
PNTMLT before and after surgery in 54 patients who improved after pudendal canal (Alcock) decompression.

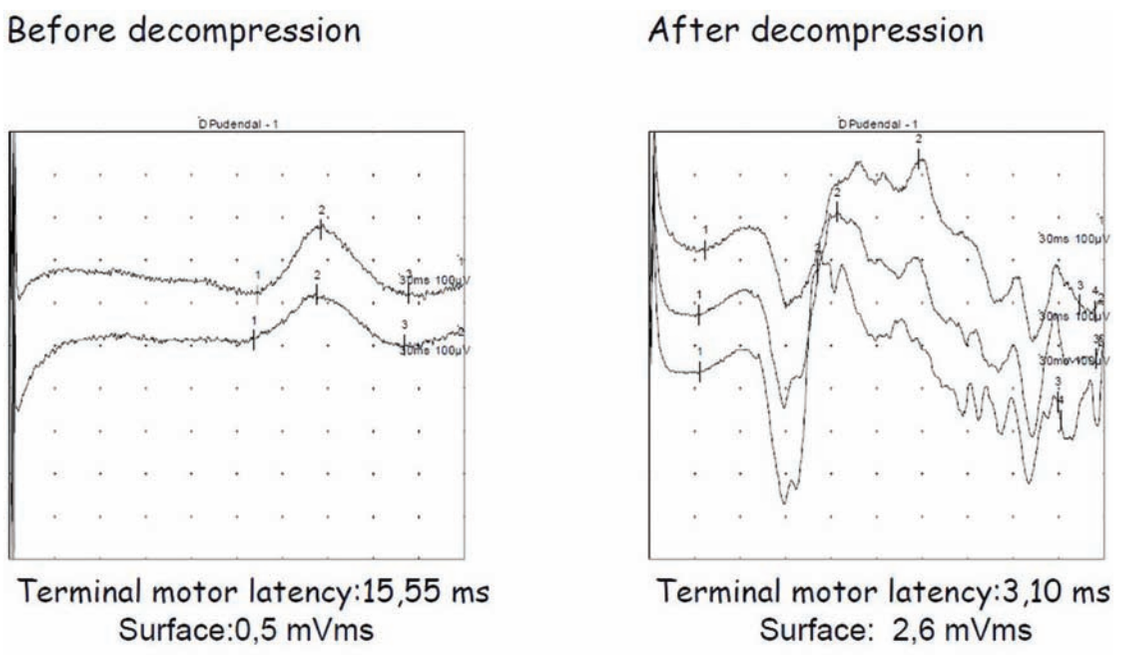

Figure 8 **Intraoperative pudendal nerve terminal motor latency test. Intraoperative recordings show that terminal motor latency decreases from 15.55 ms to 3.10 ms. The surface potential increases from 0.5 to 2.6 mVms. Changes demonstrate improvement in nerve conduction resulting from the release of a number of nerve fibers. Unpublished. Reproduced with permission from de Bisschop 2006.**

gynecologist, published about pudendal neuropathy in the 1990s. [40] Electromyography (EMG) is generally performed at the anal sphincter and the bulbocavernosus muscle. Beco and colleagues noted improvement in both the anal sphincter EMG and the PNTMLT following decompression surgery.[11] The pelvic floor muscles are relatively thin. However, neurophysiologists can develop proficiency in the testing. The innervation of the pelvic floor is complex, chiefly from the pudendal nerve, but may have additional, direct innervation from the sacral roots.[41] The obturator internus muscle may be totally or partially innervated by the pudendal nerve, but reports of EMG regarding pudendal neuropathy are unavailable.[42]

Staged sacral reflexes are assumed to localize the site(s) of pudendal nerve compression using neurophysiologic methods. It is proposed that stimulation of needles in the pubococcygeus muscle and the anterior and posterior portions of the rectal sphincter can evaluate the levels of nerve damage from the sacral spine distally.[43]

DIFFERENTIAL DIAGNOSIS

Each of the syndromes collated by the EAU may be caused by pudendal neuropathy. Publications in the literature regarding the syndromes and each of the differential diagnoses below do not define or rule out pudendal neuropathy. The simple pinprick sensory examination is not described in any clinical research articles on CPP. Only conventional morphologic and infectious causes of pelvic pain are considered. Other processes may affect the same symptoms. Neurologists typically consider herniated intervertebral disk and multiple sclerosis. Tarlov cysts are occasionally identified on MRIs. In our experience, the cysts did not contribute to CPP ($N > 20$). This is shown definitely because pudendal blocks distal to the Tarlov cyst consistently relieved the pain/symptoms. Urologists diagnose prostatitis and varicocele. Gynecologists may call the symptoms endometriosis, ovarian cyst (rupture, torsion), ovarian vein syndrome, or vestibulitis. Physical therapists diagnose myofascial dysfunctions and trigger points.

Anatomic Factors Confounding the Differential Diagnosis

Symptoms occur with pudendal neuropathy that defy typical medical concepts. Broad similarities of symptoms have significant inconsistencies. This is explainable by anatomic differences found in cadaver studies and during decompression surgery. [44] Variations occur in the origin of the pudendal nerve and its branches and in the extent of its receptive fields, and there may be congenital or acquired changes in ligaments, muscles, and bony structures [*see Table 11*].

Examples include the fascia of the obturator internus muscle (Alcock canal), which may be thin, almost not existent, or may be dense and fibrous, encasing the nerve in the canal and requiring dissection with a scalpel. A hypertrophied obturator internus muscle, seen in some athletic patients at surgery, will force the nerve medially, compressing it against the fascia of the obturator internus.

The sacrospinous ligament may be a flat structure (as typically illustrated in textbooks). The ligament may be broad or narrow [*see Figure 9*]. The nerve may be bent over a sharp edge of the ligament. The coccygeus muscle may form the bulk of the sacrospinous ligament, which consists only of small fibrous bands. The pudendal nerve may be insinuated in a tortuous pathway passing through this combined ligament and muscle structure.

The falciform process is highly variable in its breadth, thickness, and role in compression.

Extension of the ventral axial line onto the abdominal wall results in pudendal pains well beyond the base of the penis or clitoris (normal). Changes can be mapped using pinprick

Table 11 Anatomic Causes of Symptom Variability

Anatomical component	*Variations*
Pudendal neuroanatomy (congenital)[43,69,70]	Normal S2, S3, S4 nerve but prefixed or postfixed origin may include combinations from S1 to S5. Rarely L5 fibers (4% of cadavers). Nerve or branches may pass through sacrotuberous ligament, falciform process, or sacrospinous ligament. Dorsal nerve of clitoris/penis has variations of origin, exit from main trunk, pathway in Alcock canal. Inferior rectal nerve (IRN) is separate from main trunk in 10%. IRN passes through Alcock canal in only 50% of patients.
Posterior femoral cutaneous nerve (PFCN)[43]	The PFCN may be conjoined with the pudendal nerve and branch subsequently [*see Figure 13 and Figure 14*]
Bony anatomy of pelvis is remodeled (acquired), exercise, heavy manual labor[15]	Ischium extends medially. The ischial spine extends posteriorly, superiorly, and medially, changing orientation of sacrospinous and sacrotuberous ligaments [*see Figure 1b*]. Inferolateral angle of sacrum becomes broad at its base and extends laterally. Diameter of greater sciatic notch is smaller [*see Figure 1a*].
Ligament and muscle variations (congenital)	Sacrospinous ligament varies from a single structure to multiple fibrous bands. Coccygeus muscle may be the major structure between the sacrum and ischial spine. Pudendal nerve may interweave through combinations of muscular and ligamentous fibers.
Ligament and muscle variations (acquired)	Sacrotuberous ligament may become thickened and firm from repetitive trauma (cycling). Falciform process may become thick due to repetitive pivoting (basketball). Sacrospinous ligament broadens concurrent with enlargement of the ischial spine [*see Figure 1b*]. Obturator internus muscle becomes hypertrophied, compressing the nerve in the pudendal canal.
Variation of pudendal territory. Anterior: extension of ventral axial line into the thoracolumbar territory. Posterior: extension over coccyx, sacrum, and buttocks.	The cutaneous innervation by the pudendal nerve is highly variable. This can only be identified by careful pinprick examination following successful pudendal nerve anesthetic blockade [*see Figure 9, Figure 10, and Figure 11*].

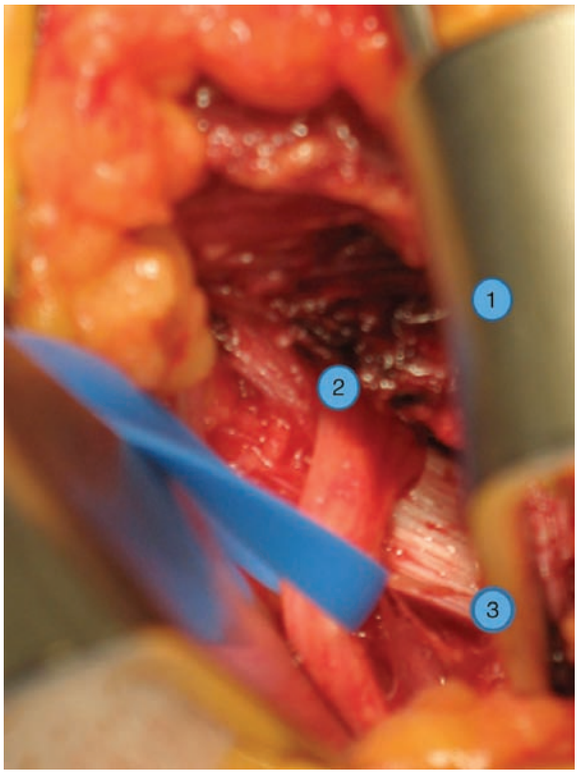

Figure 9 **Operative view: sacrospinous ligament and pudendal nerve. 1 = A retractor holds the sacrotuberous ligament open. 2 = The pudendal nerve is flat compared to distally. The nerve bends over a sharp superior edge of the sacrospinous ligament. 3 = The fibrous sacrospinous ligament is moderately narrow.**

examination after a successful pudendal nerve perineural injection (PNPI). The findings are reproducible following subsequent pudendal blocks [*see Figure 10*]. In the sacrococcygeal region, cutaneous anesthesia or hypoalgesia following a pudendal nerve block may extend high onto the sacrum, even to the waistline. Often these patients have had unsuccessful spine operations [*see Figure 11*].

Pain in the posterior thigh, neuropathy of the posterior femoral cutaneous nerve (PFCN), can accompany classic symptoms of pudendal neuropathy. This relates to anomalies of both the origin of the PFCN and a pathway medial through the pelvis [*see Figure 12*]. The PFCN may arise from the pudendal nerve, the sciatic nerve, or the inferior gluteal nerve [*see Figure 13*].[45] The PFCN may be compressed in the interligamentary space (lobster claw) along with the pudendal nerve, or it may be compressed more distally [*see Figure 14*].

Figure 15 summarizes the confounding symptoms that occur because of complex interactions of anatomical variations and physiological activities that affect the pudendal nerves.

Treatment

Effective treatments for pudendal neuropathy are similar to those of other compression neuropathies (e.g., carpal tunnel syndrome). A sequential program begins with protection and rest of the nerve (a "self-care" program) and then progresses to perineural blockades using corticosteroids and bupivacaine and, finally, decompression surgery when indicated.[10,38,46] Physical therapy is used when a high-tone pelvic floor is identified and as adjunctive therapy for the deconditioned state of many patients. Patients with "serious pain" should begin self-care and perineural injections concurrently.

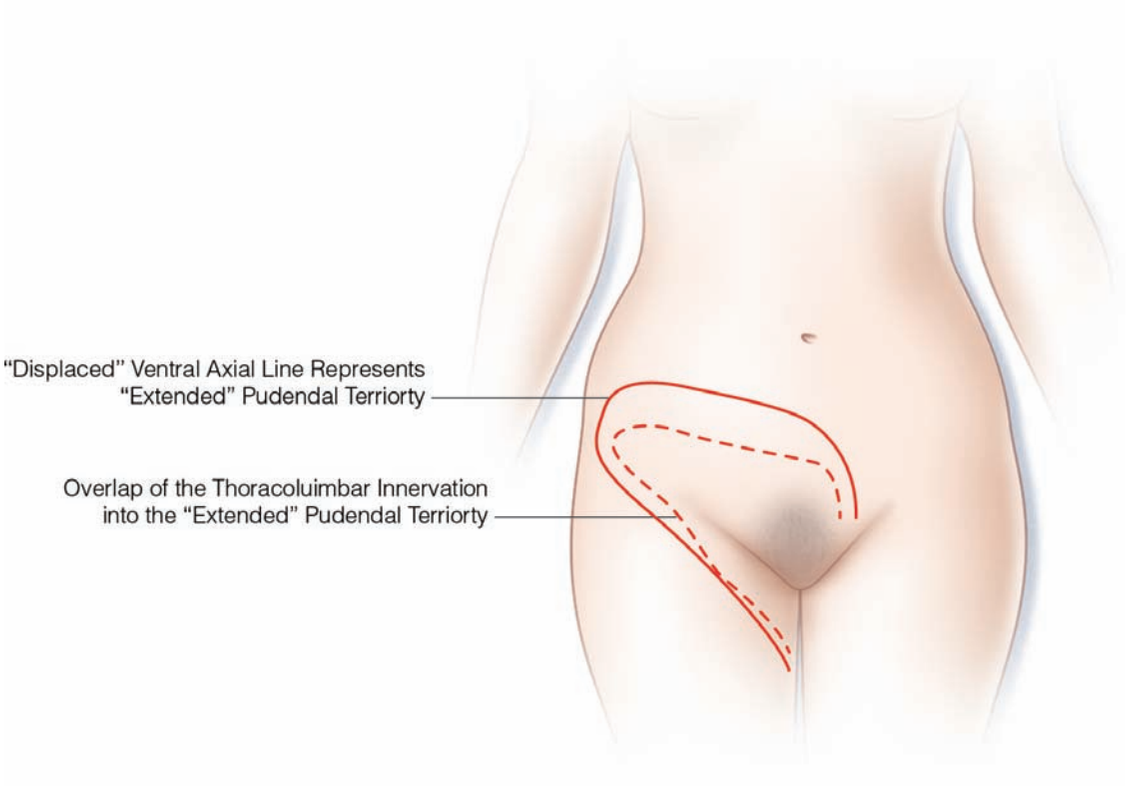

Figure 10 **Extension of the ventral axial line. A successful right pudendal nerve block relieved the chief complaint of gnawing pain in the right lower quadrant, the site of her labor pains. Pinprick analgesia extends from the clitoris, labium, and perineum to the** *solid line*, **the "displaced" ventral axial line. The normal thoracolumbar sensation overlaps into the "extended" pudendal territory and is marked with a** *dotted line* **when the pinprick started high on the abdomen or on the right lateral thigh (i.e., in the normal thoracolumbar territory). Anesthesia also overlaps the midline.**

Pain management programs incorporate integrated, comprehensive care. These address the consequences of chronic pain, including learned pain behavior and restricted socializing, depression, anxiety, and problems such as opiate hyperalgesia. The goal is to counsel toward a positive life path and limit behaviors that are associated with pain avoidance.

REST OF THE NERVE: A SELF-CARE PROGRAM

Prevention of further damage with rest of the nerve is a universal treatment and includes medications. It is effective as the only intervention in approximately 9% of patients [*see Figure 16*]. After 4 to 6 weeks, symptom scores are compared with consultation. If symptom scores confirm the patients' perceptions of poor response, then PNPIs are recommended.

The self-care program is based on patient observations that sitting on a toilet seat commonly relieved pelvic pain, as did

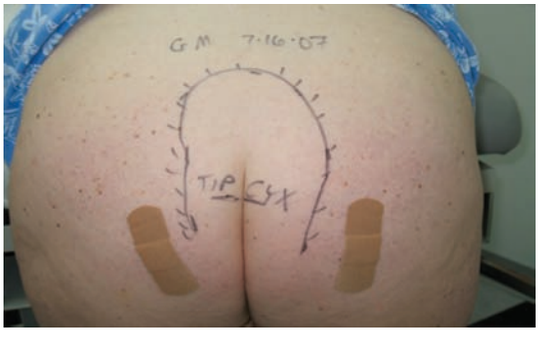

Figure 11 **Extension of the territory of the inferior rectal nerve to the coccyx (cyx) and sacrum. Analgesia following a successful pudendal nerve block (adhesive bandages) relieved the coccydynia. Pinprick examination at 2 hours after pudendal nerve perineural injection.**

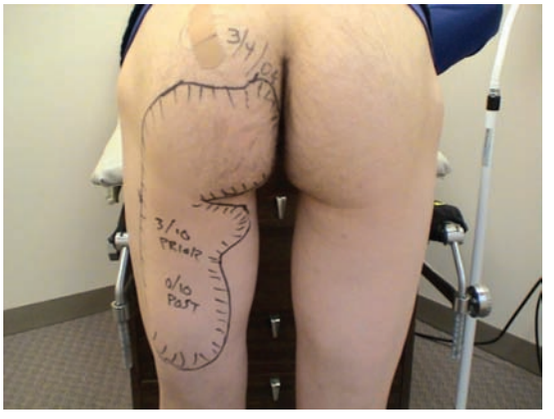

Figure 12 **Pain relief following the left pudendal block at the ischial spine (adhesive bandage). Analgesia includes the inferior rectal nerve, perineal nerve, and posterior femoral cutaneous nerve (PFCN). It is assumed that the PFCN is combined with the pudendal nerve prior to exiting the pelvis as the PFCN** [*see Figure 13 and Figure 14*]**.**

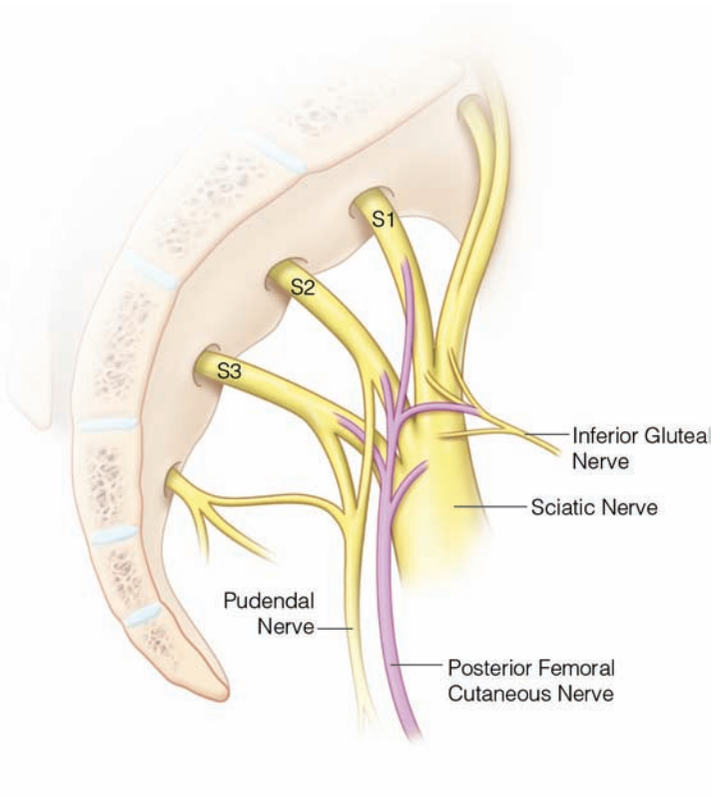

Figure 13 **Variations in the origin of the posterior femoral cutaneous nerve (PFCN). Pain distribution of PFCN neuropathy varies with the nerve origin. It may be the S1, S2, or S3 nerve; the sciatic nerve; or the inferior gluteal nerve. (after Nakanishi 1976)[45].**

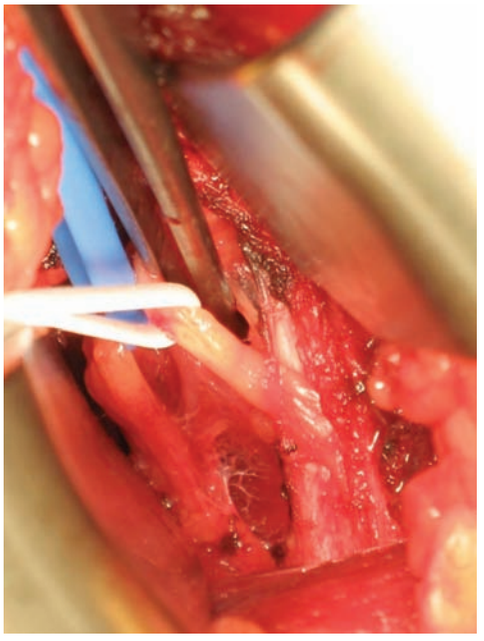

Figure 14 **Transgluteal surgery: compression of an anomalous posterior femoral cutaneous nerve (PFCN). The right pudendal nerve and PFCNs (blue vessel loops) are conjoined and compressed superiorly in the "lobster claw." After branching from the pudendal nerve, the PFCN (in the white vessel loop) is again compressed as it exits the pelvis between the sacrotuberous ligament and a thick, round falciform process.**

standing. Common contributory activities identified from patient history must be restricted or eliminated.

Self-care or nerve protection

1. Cessation of exercise activities (e.g., gymnasium machines, jogging, cycling, sit-ups) and cessation of sports activities such as hockey, soccer, and track may be required for only a short term or permanently. This can be emotionally devastating to the exercise-addicted adult. It is ruinous to high school students who anticipate athletic scholarships.
2. A standing workstation should be used by people with sedentary jobs such as computer programmers, seamstresses, and attorneys.
3. A perineal protection pad_is recommended to all patients with pudendal neuropathy. The center of the pad is cut away (like a toilet seat). Sitting on the ischial tuberosities relieves pressure on the perineum. Patients may make this pad from a gardener's kneeling pad. Commercial pads and chairs are available. Sitting on a small book or tablet with one tuberosity elevated is helpful.

Medications are prescribed for previously untreated patients: gabapentin 300 mg every 8 hours and amitriptyline 10 mg at hs, increasing every 5 days to a maximum of 50 mg. Other antieleptic, antidepressant, and sympatholytic medications may be warranted. Patients already on medications for neuropathic pain may require adjustments in therapy. We do not prescribe narcotics.

Figure 15 **Symptom variations in pudendal neuropathy.**

Symptoms may be alleviated by topical compounded creams. Various combinations can be ordered (e.g., gabapentin + tetracaine + diazepam; bupivacaine + clonidine + ketamine). Compounded rectal or vaginal suppositories using similar mixtures are often beneficial.

Patients are encouraged to avoid foods containing aspartame sweeteners and to use a polyamine-light diet. Each of these compounds may aggravate pain in CPP patients.

Self-care is durable as long as 14 years in our experience.

PUDENDAL NERVE PERINEURAL INJECTIONS

Perineural infiltration of local anesthetics and corticosteroid medication is recommended for 90% of patients who fail the self-care and medications. Bensignor and colleagues were successful using a series of three PNPIs at 4-week intervals.[47] Our experience confirms this protocol while monitoring weekly symptom scores for 3 months. A single PNPI is rarely curative. After a PNPI, there is an immediate response to bupivacaine followed by more durable pain relief beginning approximately 2 to 10 days after the block, which maximizes at about 3 to 4 weeks. If subsequent PNPIs are delayed, the pain gradually worsens. PNPIs use 6 mL of 0.25% bupivacaine and 1 mL of methylprednisolone (40 mg), triamcinolone, or betamethasone. Two PNPIs are given into the interligamentary space medial to the ischial spine (the major site of nerve compression) [*see Figure 17*]. The third PNPI, into the Alcock canal, is performed by an interventional radiologist using CT guidance for needle placement. Examination of the six pudendal nerve branches 2 hours after each pudendal block determines the "quality" of the nerve block. Pain control correlates directly with the number of nerve branches that are anesthetized.[24] Treatment of any "perimeter neuropathies" is performed following the PNPI. These painful secondary neuropathies may require weekly injections to relieve the secondary neuritic

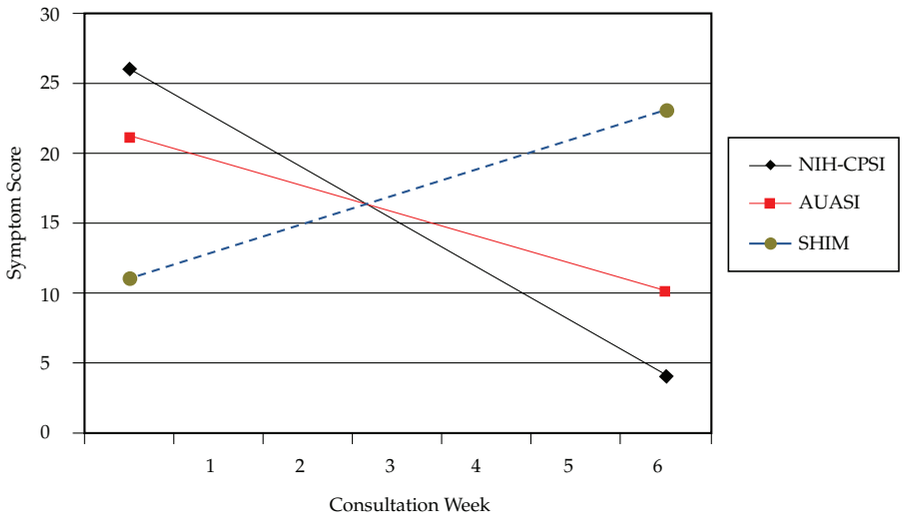

Figure 16 **Response to self-care treatment (nerve protection) and amitriptyline. Pains in this 35-year-old man had not responded over many months to multiple medications and implantation of a sacral nerve root stimulator. All symptom scores improved after 6 weeks of self-care and amitriptyline 50 mg at hs. He was able to turn off the neuromodulation device. AUASI = American Urological Association Symptom Index; NIH-CPSI = National Institutes of Health Chronic Prostatitis Symptom Index; SHIM = Sexual Health Inventory for Men.**

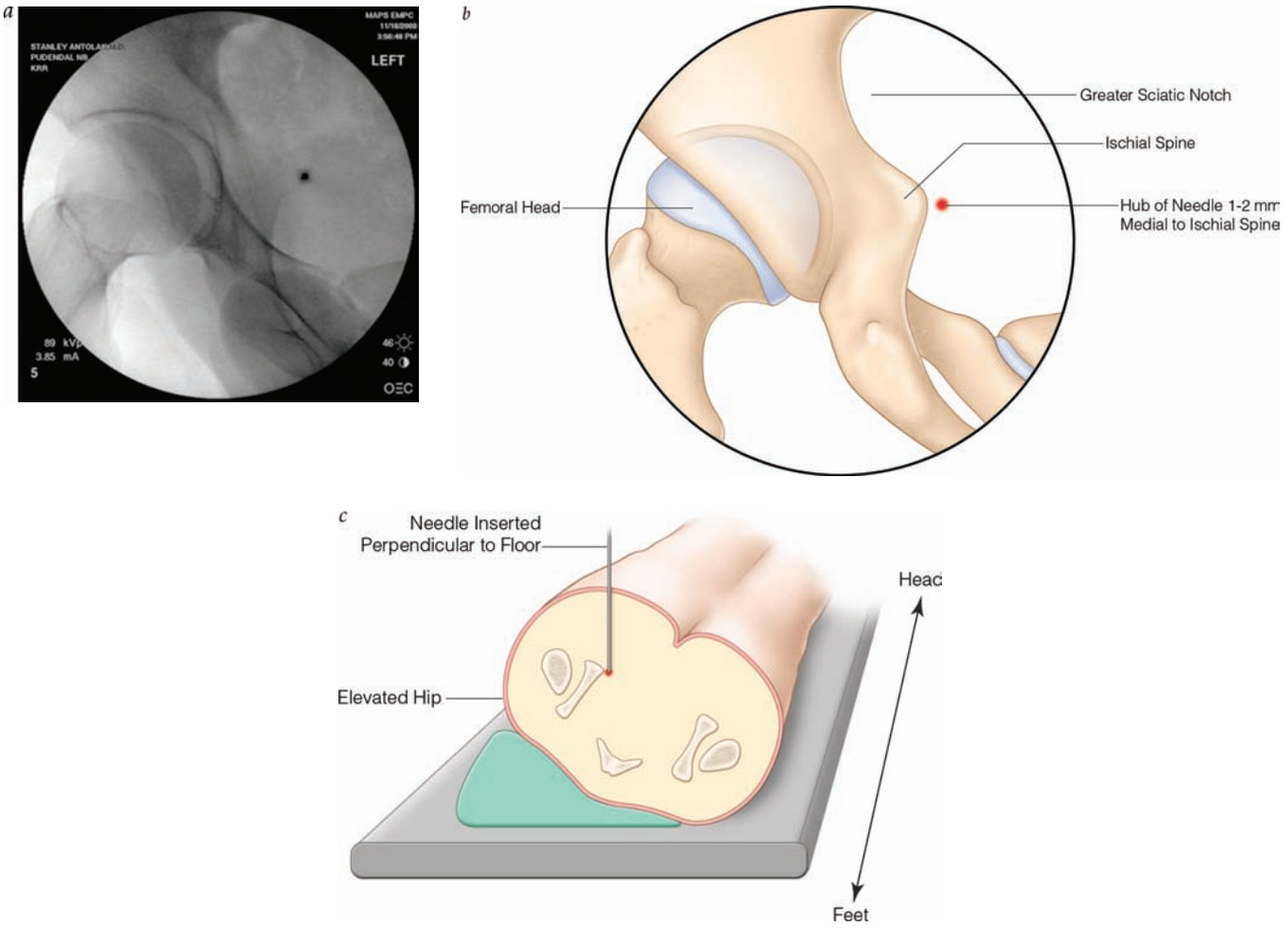

Figure 17 **Fluoroscopy-guided pudendal nerve perineural injection (PNPI) into the interligamentary space ("lobster claw"). (a) Hub of the needle medial to the ischial spine. (b) 1 = femoral head; 2 = ischial spine with the hub of the needle medially 1 to 2 mm; 3 = greater sciatic notch. (c) Position for PNPI: the ipsilateral hip is elevated. The needle is inserted perpendicular to the floor.**

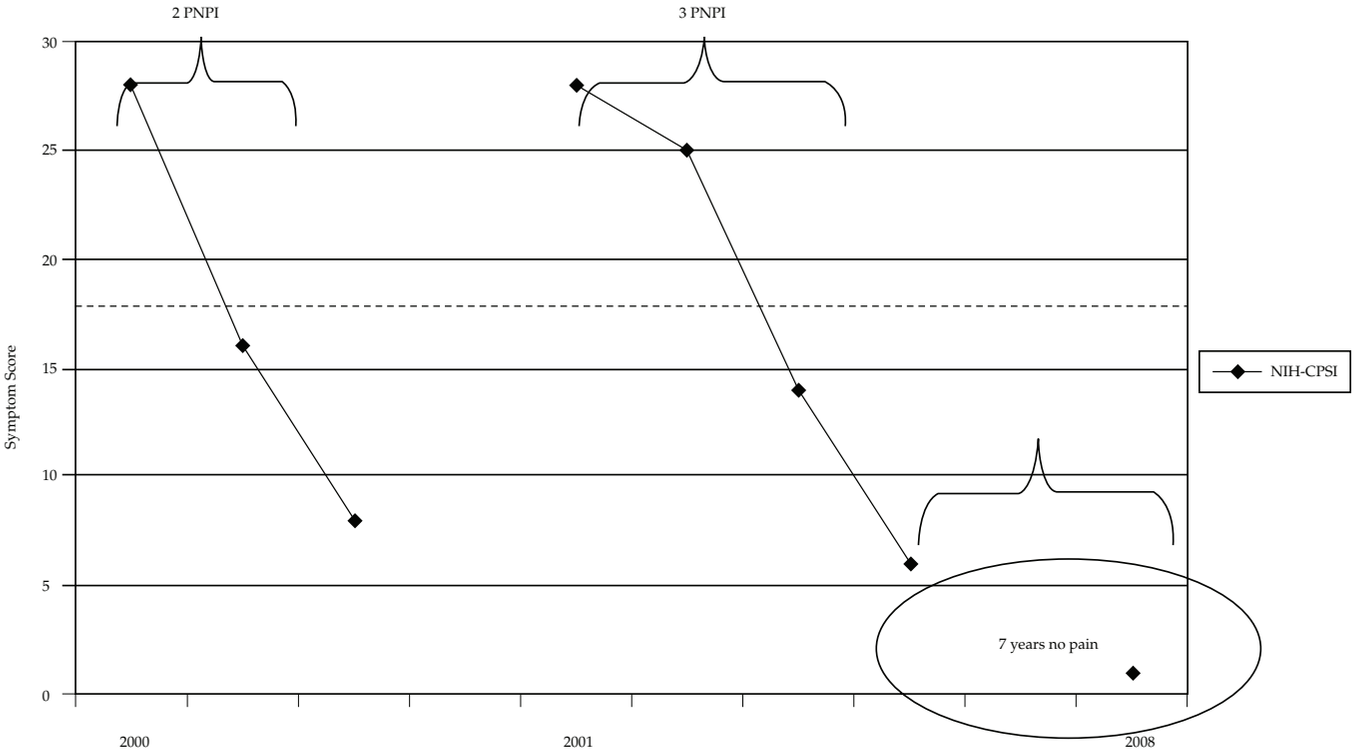

Figure 18 **Long-term response following a second series of pudendal nerve perineural injection (PNPI). A 78-year-old pilot with perineal and scrotal pain became pain free after two PNPIs in 2000. Pain recurred and required three PNPIs in 2001. He remained pain free when contacted in 2008. The x-axis intervals are 4 weeks. A score of 18 or more is abnormal. NIH-CPSI = National Institutes of Health Chronic Prostatitis Symptom Index.**

pains and control central sensitization. A physical therapist can assist with postural correction exercises.

Improvement and cures following PNPI are durable as long as 13 years, measured by symptom scores. Interval PNPIs may be needed for recurrent pain after 3 months to several years [*see Figure 18*]. One or more repeat series of three PNPIs may be necessary to provide long-term symptom control**.** About 35% of patients fail the conservative treatments and become candidates for decompression surgery.

The medical literature discussing PNPI is challenging because of inconsistencies; techniques vary, early response to PNPI is often not recorded, follow-up intervals are not uniform, only one injection may be offered, and only lidocaine may be injected. Needle placement using a nerve stimulator may produce more consistent responses and needs study.[48] Failures occur because ligaments, fascias, and hypertrophied muscles are inconsistent among patients, thus affecting placement of medications. [49]

PNPI should not be considered a diagnostic tool for pudendal neuropathy. Failure of pain control occurs in up to 32% of injections in published series. A definite diagnosis can be made clinically in over 90% and increases to 100% using neurophysiologic testing in suspected cases. PNPIs are a therapeutic method, not a diagnostic tool.

Pulsed radiofrequency is being used with some benefit.[50] Injection of botulinum toxin into pelvic floor muscles has been recommended. These require controlled studies after a definite diagnosis of pudendal neuropathy has been made.

SURGICAL DECOMPRESSION OF THE PUDENDAL NERVE

Decompression surgery is the only method to diagnose pudendal nerve entrapment and may relieve or reduce symptoms. The transgluteal approach is the most common method for decompression of the pudendal nerve. It offers direct vision of the nerve pathway from the proximal to the ischial spine almost to the urogenital diaphragm. Bilateral surgery is necessary in over 90% of the patients. After unilateral surgery, one half of the patients will request contralateral surgery. Transvaginal, perineal, and laparoscopic approaches are performed by several authors.[11,39,51,52] Antalgic surgery, used to limit windup, includes an epidural anesthetic, intravenous ketamine, and infiltration of local anesthetics at the incision.

Surgery may be performed as an outpatient or an inpatient for 1 to 2 days. A prone, flexed position is used [*see Figure 19*]. Gluteus muscle fibers are separated. The sacrotuberous ligament is opened longitudinally followed by blunt dissection of the ischiorectal fossa. The pudendal nerve is identified, and all compressing ligaments and fibrous bands are divided. The nerve is transposed medially and anterior to the ischial spine (analogous to transposition of the ulnar nerve). An elongated ischial spine may require excision. Hyaluronidase film, amniotic membrane, and α_2-macroglobulin are useful for the prevention of perineural adhesions. Bupivacaine can be administered for several days using a percutaneous catheter. Preexisting central sensitization must be addressed throughout the postoperative period because of windup precipitated by the procedure. This may require bupivacaine epidural analgesia and intravenous ketamine.

Table 12 Reasons for Failure of Surgery to Control Pain

Problem	*Diagnosis*	*Treatment*
Serious compression of pudendal nerve	Observation during surgery	PNPI [*see Figure 18*], medications, IV ketamine, epidural bupivacaine infusion
Sympathetically maintained pain	Difficult; observe for skin changes over natal cleft	Oral clonidine, lumbar or presacral sympathetic blockade
Central sensitization	Persistent organ dysfunction (visceral reflexes)	PNPI, medications, IV ketamine, epidural bupivacaine infusion
Secondary pain generators	Evaluation at consultation is repeated postoperatively	Anesthetic infiltrations, physical therapy, topical compounds, ilioinguinal neurectomy [*see Figure 25*]

IV = intravenous; PNPI = pudendal nerve perineural injection.

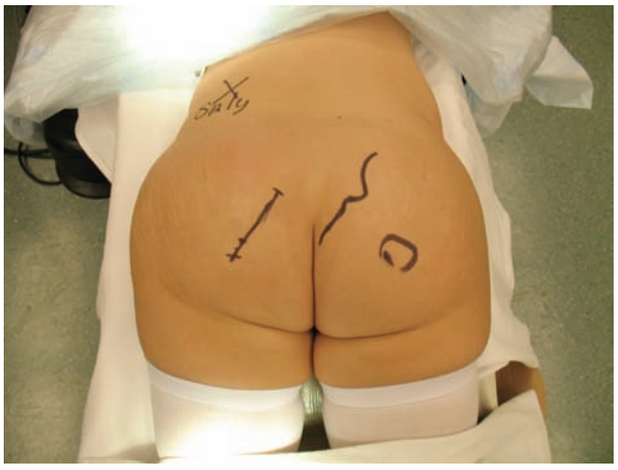

Figure 19 **Incision site for left unilateral transgluteal decompression surgery. The patient is in a prone, flexed position. The midline of the left sacrotuberous ligament is marked. The right ischial tuberosity and sacrum with a prominent inferolateral angle are outlined as reference points.**

Operative Results: Postoperative Pain

Pain relief is associated with the duration and severity of symptoms preoperatively and the duration of pain control following a series of preoperative PNPIs. Published results suggest that 62 to 75% have a cure or good response to decompression surgery [*see Figure 20*]. Pain relief following decompression may be immediate, but more often pain decreases over 9 to 24 months. Patients attest to continuing improvement over 3 to 7 years. The only randomized controlled study of transgluteal decompression surgery showed that statistical improvement persisted at 48 months.[53] Bladder, bowel, and sexual function can normalize rapidly or require many months. Standard scores have not been used for comparisons. Severe compression noted during surgery and other factors explain persistent postoperative pain [*see Table 12*].

Postoperative pudendal pain may respond to a series of weekly pudendal nerve blocks using steroids, heparin, and bupivacaine [*see Figure 21*]. Early, unpublished reports by Dr. Dimitri Chagava (2012) at the Burdenko Neurosurgery Insti tute in Moscow indicate success using pulsed radiofrequency

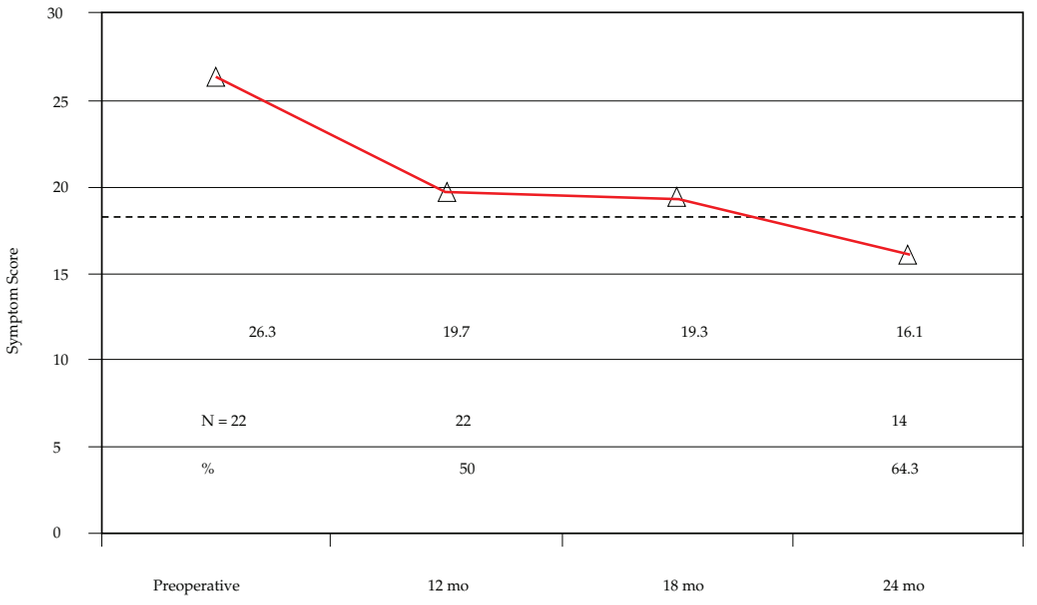

Figure 20 **Responses to surgery. Cumulative averages of symptom scores in 22 men (31.4% of this treatment cohort required surgery). Fifty percent were improved at 12 months and 64.3% were improved at 24 months. *N* = number of men in the postoperative time group; % = percentage of men responding to surgery.**

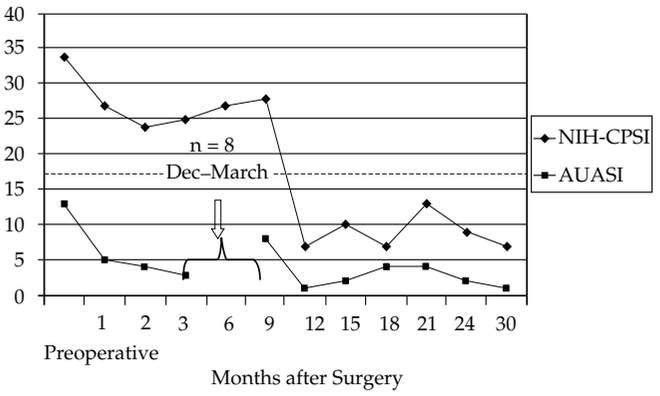

Figure 21 **Surgical "failure" RX post-op Heparin PNPI ↓ n = 8. Pudendal nerve perineural injections (PNPIs) provide complete postoperative pain control. Male: 8 weekly PNPIs beginning 3 months postoperatively using heparin, bupivacaine and betamethasone. No steroid was injected at weeks 2, 3, 4, 6, 7, and 8. AUASI = American Urological Association Symptom Index; NIH-CPSI = National Institutes of Health Chronic Prostatitis Symptom Index.**

without steroids for control of persistent postoperative pain. Sacral neuromodulation is not Food and Drug Administration approved, and if used after decompression surgery fails, sacral neuromodulation should be delayed for 24 months to permit normal healing. Preoperative medications should be continued or adjusted. It is imperative to treat the secondary neuropathic pain generators.

Complications

The self-care/rest program has few complications. These relate to the standing and other postural adjustments required to prevent compression by sitting. Patients often have aching feet and legs.

Following PNPI, there are a few problems, estimated at less than 1%. Pain flares may occur following initial analgesia. Onset is usually after a few days and lasts several days. Bupivacaine may spread to the sciatic nerve, causing transient leg weakness. Intravenous injection is rare and has a transient effect on mentation, pulse, and blood pressure lasting only a few minutes. In my experience with several thousand PNPIs, no patients required emergency interventions. The most serious complication is penetration of the nerve by the injection needle. There is immediate severe pain that may persist for 6 weeks or longer.[46]

Procedural complications during or following surgery include transection of a branch of the nerve, requiring repair in less than 0.1% of operations. The wound infection rate is less than 1%.

Persistent pain is not a complication because the severity of intrinsic nerve damage cannot be evaluated. Most surgeons recognize a plateau in the rate of pain reduction between 6 and 12 to 18 months. The reason for this phenomenon is unknown; however, patients often resume "normal" exercise activities too early and may redamage the nerve. Secondary pain generators require treatment. Sympathetically maintained pain requires treatment. After excision of the sacrospinous ligament, the nerve is still susceptible to physiologic compression between the pelvic floor and the sacrotuberous ligament while sitting.

Destabilization of the pelvis occurred in one patient when surgery included transection of both the sacrotuberous and sacrospinous ligaments. The procedure is now performed through a longitudinal incision in the sacrotuberous ligament.

Measuring Treatment Results; Quality of Care

Postoperative neurophysiologic test comparisons are not reported frequently; however, both anal sphincter EMG and PNTMLT improve [*see Table 8*].[54] The WDT may normalize after pudendal decompression surgery.[19] Benson and McClellan identified worsening PNTMLT results following vaginal gynecologic operations.[55]

Validated symptom scores are widely accepted as evidence of treatment responses. At present, there is no standardization or coordination among clinical researchers or in clinical practice. Since 1998, the following scores, which are available in several languages, have provided information on individual patients and permit comparisons of treatment groups:

1. The NIH-CPSI[7] queries pelvic pain and some voiding complaints. It has four domains: pain, urination, impact of symptoms, and quality of life. Exchanging female anatomic terms permits effective use in females.
2. The AUASI[56] or International Prostate Symptom Score (IPPS) measures urination symptoms, not prostatism, and has been validated in females.
3. The SHIM[57] is a brief evaluation of sexual function containing five questions.
4. The Female Sexual Function Index (FSFI)[58] is available. but women complain that it is tedious and intrusive.

Uniformity has not been achieved in adopting questionnaires regarding neuropathic pain. The IASP reviewed a number of indices useful for both clinical and research use in patients with chronic pain.[59] Questionnaires are also published for associated symptoms, such as irritable bowel and interstitial cystitis.

The Collaborative Health Outcomes Information Registry (CHOIR) is an effective database that may become the best method of retaining and retrieving patient data, as demonstrated by Stanford University researchers. At present, it is not easily incorporated into most electronic medical records.[60]

Prognosis

Patients who respond to self-care or PNPI may have recurrent symptoms intermittently. The anatomic pathway remains unchanged, and the nerve is at risk for future injury, often caused by hip flexion activities. Approximately 35% of patients with definite pudendal neuropathy require decompression surgery. Averages of symptom scores are normal by 24 months [*see Figure 22*]. Pain relief after surgery is slow [*see Figure 23*]. Durable cures of serious CPP in patients treated at the Center for Urologic and Pelvic Pain are as long as 9 years using self-care, 13 years after pudendal nerve blocks, and 12 years after decompression surgery. Many patients will live with some symptoms. International reports indicate that decompression surgery fails to provide a "good response" in 25 to 35% of patients and may require continuing medications. Patient attestations

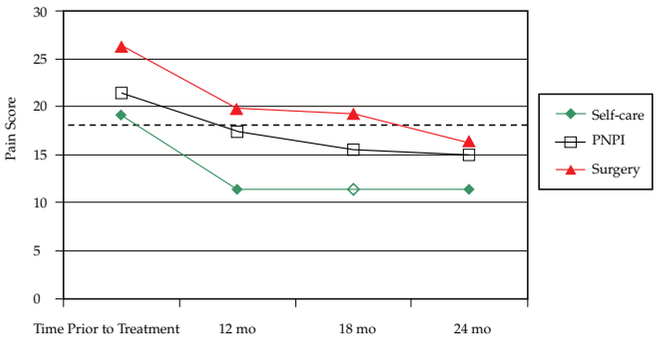

Figure 22 **Results of three successive treatments for pudendal neuropathy. The National Institutes of Health Chronic Prostatitis Symptom Index (NIH-CPSI) responses in 70 men treated sequentially are shown. A score greater than 17 is abnormal. Note that each group has increasingly higher pain scores and requires a progressively longer time for a reduction in symptoms. PNPI = pudendal nerve perineural injection.**

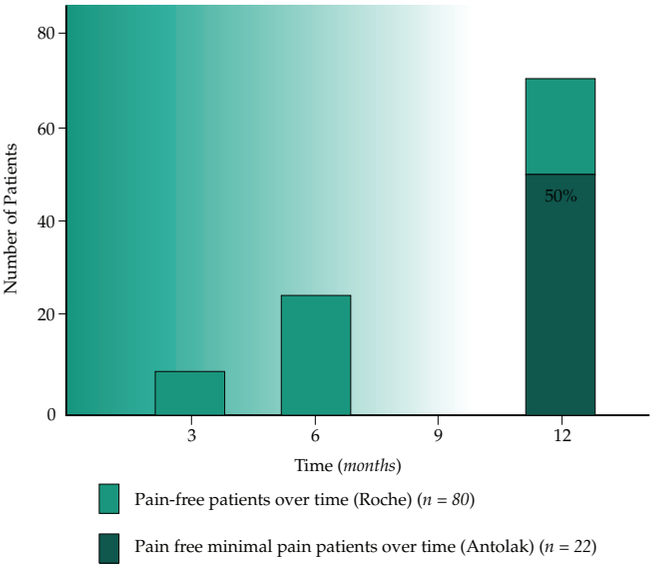

Figure 23 **Postoperative surgical results.[68] The results are recorded as pain-free status. Standard scores were not recorded.**

indicate slow improvement as late as 3 to 5 years after nerve decompression. Suicide has occurred—definitely in one patient (gunshot) and possibly in three (alcohol and drug overdose).

Painful Neuropathies in the Perimeter of the Pudendal Territory

Sixty-four percent of our patients have identifiable, treatable additional or secondary or "perimeter" neuropathies affecting or overlapping into the pudendal territory [*see Table 7*]. Concurrent diagnosis and treatment of these vexatious pain generators are necessary to resolve patients' pelvic pains.[*see Figure 24*]

The thoracolumbar junction syndrome (posterior ramus syndrome, Maigne syndrome)[62] is found in 57.6% of females versus 11.6% of males with pudendal neuropathy. It affects one or more levels from T6 to L2. It is suspected that the posterior rami are repeatedly stretched as they pass through the fascia of the multifidus/sacrospinalis muscle. Patients have a slouched posture from work over a computer or heavy manual labor or performing excessive exercises requiring flexion of the back and waist. Young girls learn to sit in a hunched position to avoid embarrassment when teased in middle school and high school about breast development. The antalgic position is a major cause. Irritated posterior rami may cause abdominal pain and tenderness and also visceral complaints that mimic irritable bowel syndrome or painful bladder syndrome. Diagnosis is clinical

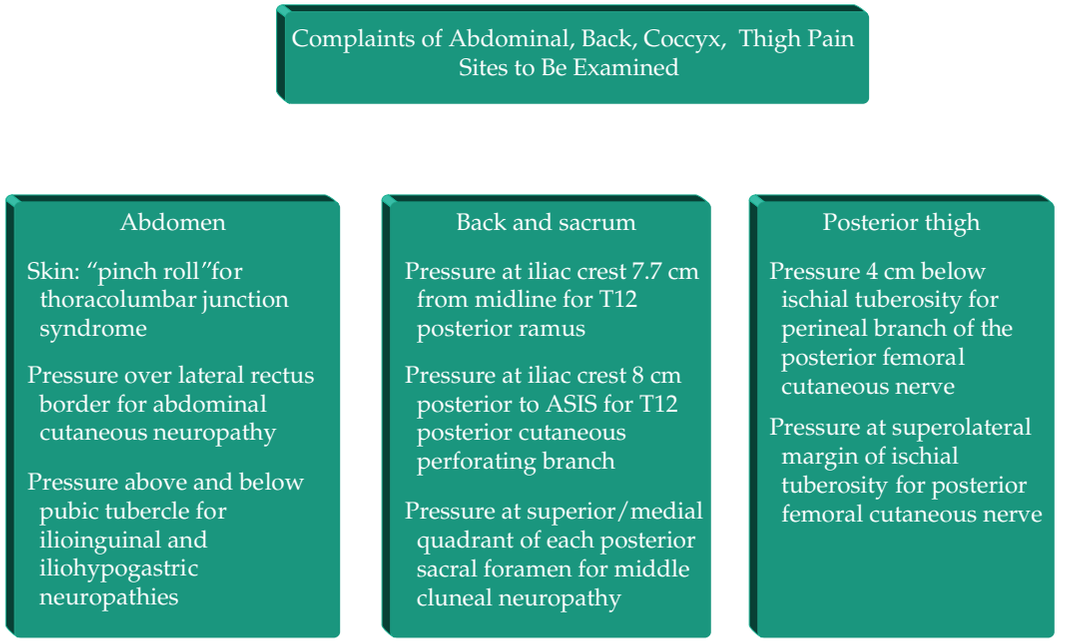

Figure 24 **Physical examination for secondary or "perimeter" neuropathic pain generators. ASIS = Anterior superior iliac spine.**

using a "pinch roll test" from the flank to the pubis with the patient in the supine position [*see Figure 25*]. Painful pinch roll is pathognomonic for thoracolumbar junction syndrome. Pressure on the spinous processes with the patient flexed over the examination table produces tenderness at those sites, but the pains are not referred anteriorly. Postural correction is imperative to straighten the posture of the thoracic spine. Infiltration of the subcutaneous tissues of the abdominal wall with a mixture of 0.25% bupivacaine and 1% lidocaine will provide immediate and often prolonged relief [*see Figure 26*]. Some patients find that superficial paraspinal blocks 1 cm from the midline may provide better relief of abdominal pain and symptoms in some patients [*see Figure 27*]. Weekly blocks may be required early, followed by monthly blocks for several months. During this time, a postural correction program appears to be most beneficial.

ABDOMINAL CUTANEOUS NEUROPATHIES

The medial branches of the anterior rami from T6 to L2 may cause both pain and reflex somatovisceral symptoms, including irritable bowel syndrome and symptoms suggesting interstitial cystitis (painful bladder syndrome). The etiology appears to be a stretch, compression, and irritation of the nerves as they pass through abdominal wall fascias and is related to zealous exercising.[63] Diagnosis is made by pressure at the lateral border of the rectus muscle. Pain is usually felt locally under the fingertip. However, referred pain may go to the rectum, penis, or clitoris labia or vagina and even into the leg. The examination may cause nausea or an urge to void.

Treatment is local infiltration of bupivacaine/lidocaine at each tender nerve at the lateral border of the rectus muscle. Postural correction is recommended and is helpful. Frequent infiltrations may be necessary. Corticosteroids are not used because most patients are having concurrent treatment of pudendal neuropathy with those medications. One patient with Crohn disease responded after two injections for pudendal neuropathy and multiple abdominal cutaneous neuropathies. Diarrhea ceased, stools became normal (one daily), pain was minimal, and she was able to return to work for the

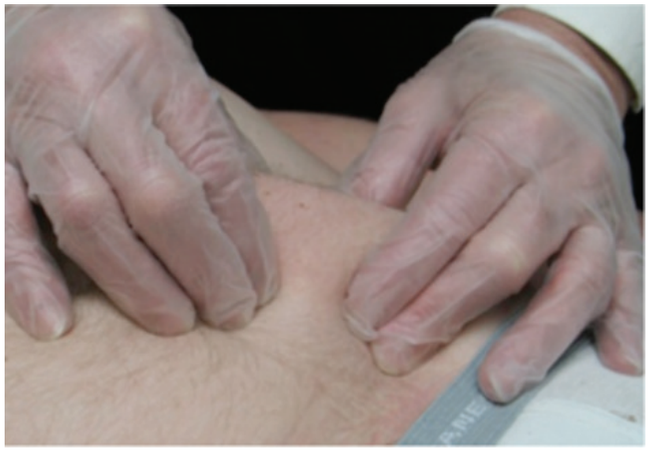

Figure 25 **Pinch roll test for posterior ramus (Maigne) syndrome. The skin is lifted, slightly pinched, and rolled. Begin at the flank and proceed medially and inferiorly to the pubis. The test is also effective over the greater trochanter for T12 posterior ramus neuropathy.**

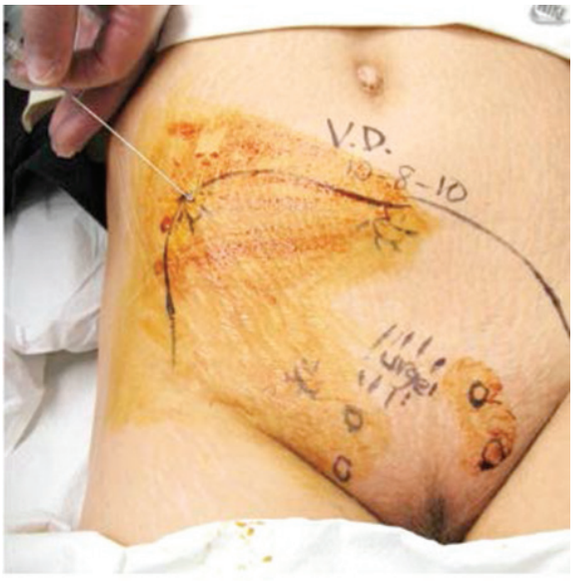

Figure 26 **Abdominal wall injections for posterior ramus syndrome. A large painful area of the pinch roll test is outlined by a *solid line*. One small area caused the urge to void and is outlined by *hash marks*. Treatment was subcutaneous infiltration of 48 mL bupivacaine/lidocaine using a 12 cm 22-gauge needle. Note that concurrent ilioinguinal and iliohypogastric neuropathies also require infiltrations. All pains were relieved. Future blocks will be necessary while the patient continues postural correction exercises.**

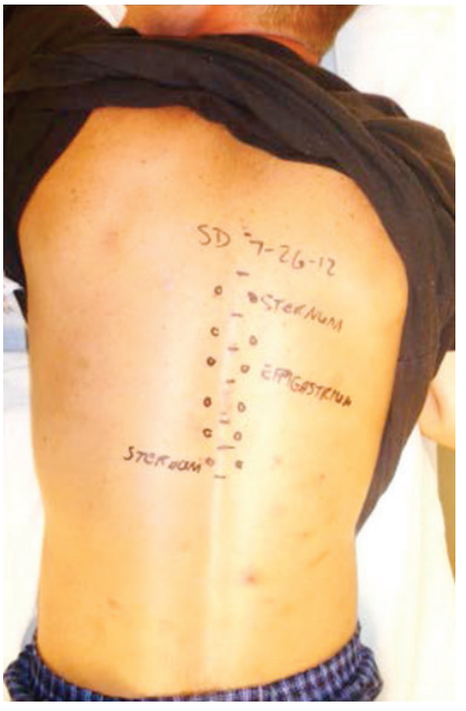

Figure 27 **Subcutaneous paraspinal blocks of the posterior rami (after Maigne). The needle is placed 1 cm lateral to the midline at the level of the spinous processes. One to 4 mL of bupivacaine/lidocaine is infiltrated at each site. Some patients find better pain relief using this method. Needle stimulation of posterior rami generators causes epigastric and sternal pains in this man with nausea and vomiting following bowel movements.**

first time in 6 years. She had undergone five colon operations because of cramping, diarrhea, and weight loss.

Abdominal cutaneous neuropathies were identified in 15.8% of females and 5.4% of males in a patient care review.

ILIOINGUINAL AND ILIOHYPOGASTRIC NEUROPATHIES

Ilioinguinal and iliohypogastric neuropathies occur in a complex region of overlapping neuropathies in the suprapubic, inguinal, and scrotal/labial regions.[64] However, in most patients, the diagnosis can be made by pressure over each nerve above and below the pubic tubercle. The pain can be life altering because of its persistence and repetitive misdiagnosis. Inguinal hernia repair, either laparoscopic or open with or without placement of mesh, is a common cause. A Pfannenstiel incision (cesarean section) may damage the nerves at the lateral extent. Our experience indicates that ilioinguinal and iliohypogastric neuropathies developed related to job activities such as lifting, carrying, and twisting with loads or following exercise activities.

Treatment includes rest, postural correction exercises, and infiltration of bupivacaine/lidocaine above and below the pubic tubercle or 2 cm medial to the anterosuperior iliac spine. A transversus abdominis plane (TAP) block using local anesthetics may relieve the pain for a short time.[65] Repeat TAP block may slowly resolve the pain. Cryoablation may provide several weeks to several months of pain control but may be completely unsuccessful. Ilioinguinal and iliohypogastric neurectomies can provide complete relief in over 90% of cases and may be necessary when associated with pudendal neuropathy [*see Figure 28*]. Neuromas have been found. Entrapment of the nerve has occurred from suturing a laparoscopy port.

In our patient population with pudendal neuropathy, unilateral ilioinguinal/iliohypogastric neuropathies occurred in 35.3% of males and 11.5% of females. Bilateral ilioinguinal and iliohypogastric neurectomies occurred in 23.5% of males and 38.4% of females.

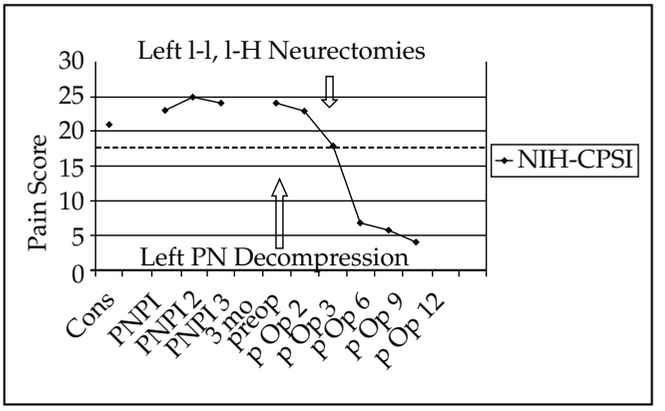

Figure 28 **Postoperative pain control required ilioinguinal neurectomy. Pudendal pain was eliminated after pudendal decompression. Longstanding ilioinguinal neuropathy persisted. Complete pain relief was achieved after left ilioinguinal neurectomy. I-H = iliohypogastric; I-I = ilioinguinal; NIH-CPSI = National Institutes of Health Chronic Prostatitis Symptom Index; PN = pudendal nerve; PNPI = pudendal nerve perineural injection.**

T12 POSTERIOR RAMUS NEUROPATHIES

T12 posterior ramus neuropathies usually occur together [*see Figure 29*]. The posterior ramus can be assessed using pressure at the iliac crest 7.7 cm from the midline.[66] This will reproduce low back and buttock pain. Pains may be referred to the T12 territory anteriorly at the lower quadrant. Most patients respond to simple treatment of bupivacaine/lidocaine with or without corticosteroids. Multiple injections over a period of weeks may be necessary. Concurrently, patients continue a postural correction exercise program for the thoracic spine. The incidence and prevalence have not been recorded in our practice. It occurs quite frequently.

Neuropathy of the cutaneous perforating branch of the posterior ramus also occurs rather frequently [*see Figure 30*]. Patients have complaints of low back, buttock, and hip pain. The diag-

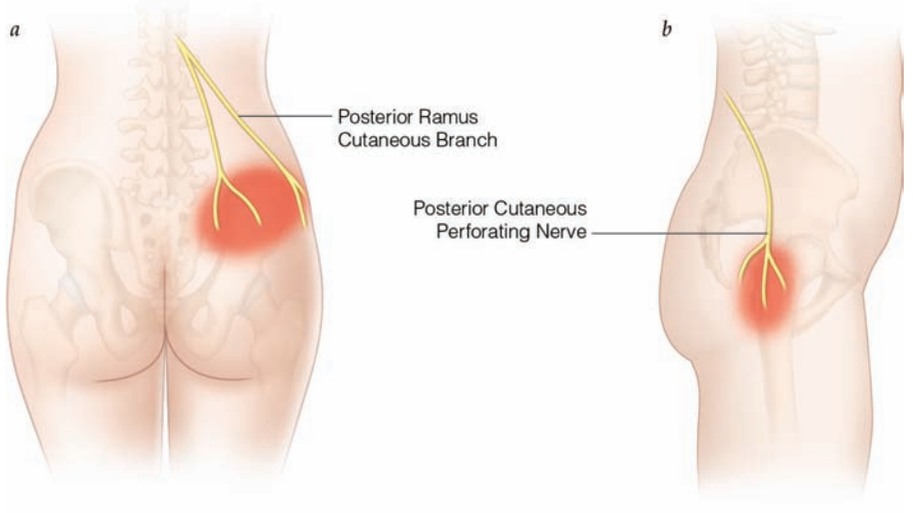

Figure 29 **Distribution of T12 posterior rami. Complex pain in the inguinal region, low back, and "hip." Painful skin rolling may occur over the greater trochanter.**

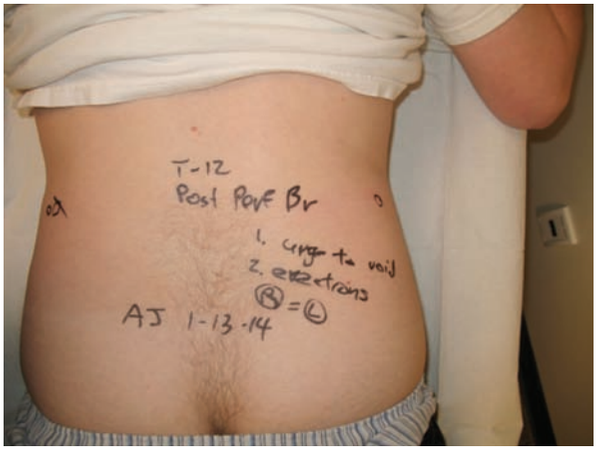

Figure 30 **Neuropathy of the T12 posterior cutaneous perforating branch. Penile pain, unwanted spontaneous erections, and urge to void were reproduced by pressure laterally at the iliac crest over the pathway of the T12 posterior cutaneous perforating branch.**

nosis is made using pressure over the iliac crest approximately 8 cm posterior to the anterior superior iliac spine, the lateral crestal point. Painful skin rolling over the greater trochanter may be found. Cellulalgia, a tender thickening of the skin similar to peau d'orange, may be present at this site. Treatment is infiltration of the nerve with bupivacaine/lidocaine and postural correction exercises for the thoracolumbar junction.

MIDDLE CLUNEAL NEUROPATHY

Middle cluneal neuropathy (MCN) is a vexatious neuropathy causing low back and buttock pains.[65] When undiagnosed, it often results in multiple failed back surgeries, including intervertebral disk and fusion operations. There are few reports of this neuropathy in the medical literature. MCNs are found in 35.3% of males and 56% of females with pudendal neuropathy. The cutaneous distribution is chiefly over the buttocks. The cutaneous branches of S2. S3, and S4 posterior rami appear to be trapped or "irritated" as they pass through the multifidus/sacrospinalis muscle fascia. About 30% are associated with a fatty herniation through this fascia, an episacroiliac lipoma or "back mouse."[68] Forward flexion causes low back and buttock pain (brushing teeth, washing dishes). The MCN and pudendal nerves originate from common spinal cord levels. Patients with MCN may have associated bladder, bowel, or sexual complaints independent of pudendal neuropathy. MCN may also be associated with lower quadrant abdominal, perineal, genital, and hip pains. The middle cluneal nerves provide a branch to the sacroiliac joint, which may lead to erroneous diagnosis of sacroiliac joint disease.

Treatment of MCN begins with avoiding bending and lifting. Persons with short legs (knee to foot) are encouraged to use a 4-inch lift when sitting. Airplane seat design often aggravates the low back pain of MCN. Infiltration of bupivacaine/lidocaine mixture will relieve pains for variable durations [*see Figure 31*]. The addition of corticosteroids prolongs pain control for several weeks. Topical compounded creams (e.g., neuroleptic, tetracaine) may also decrease pain and reduce central sensitization.

Cryoablation or radiofrequency ablation of these nerves has been considered, but there are no statistics. Surgical excision of the back mouse is occasionally helpful. There is typically an enlargement of the fascial fenestra for the nerve. Excision of a short segment of the nerve has proved the most effective surgical treatment [*see Figure 32*].

NEUROPATHY OF THE PERINEAL BRANCH OF THE PFCN

Neuropathy of the perineal branch of the PFCN is often identified in women with pain near the introitus or anus and extending into the crural fold toward the base of the clitoris (scrotum in men). Pain may be associated with persistent genital arousal

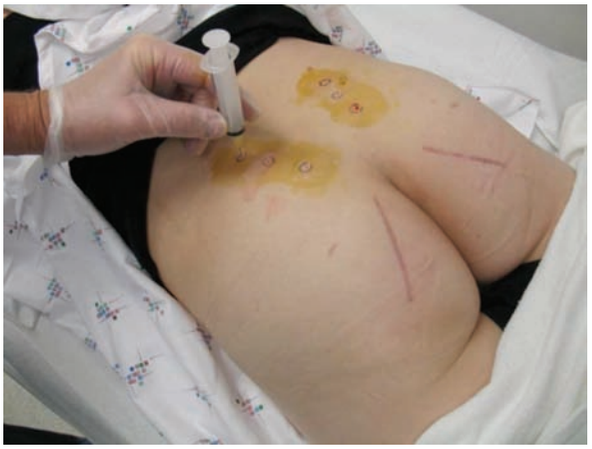

Figure 31 **Injection of the S2, S3, and S4 middle cluneal nerves bilaterally. This woman had low back pain persisting after pudendal decompression** (*scars*)**. Blocks were made medial and superior to the sacral foramina. Pain was relieved after repeat blocks with steroid/bupivacaine and concurrent postural correction exercises. This left-side injection is made near the 10 o'clock position (upper outer quadrant) of the S2 foramen. A mixture of medications is infiltrated when the needle touches sacral bone.**

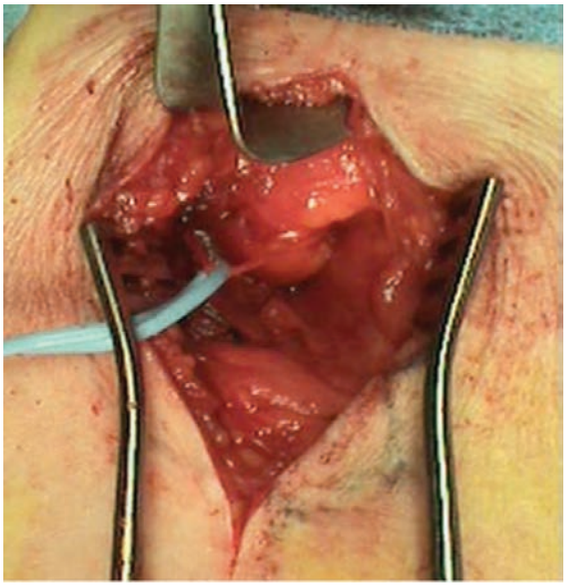

Figure 32 **Middle cluneal neuropathy with low back pain. Neurectomy. An episacroiliac lipoma distorts the pathway of the middle cluneal nerve (in the vessel loop). The lipoma and a 1 to 2 cm portion of nerve will be excised. The fascial defect will be closed with mesh.**

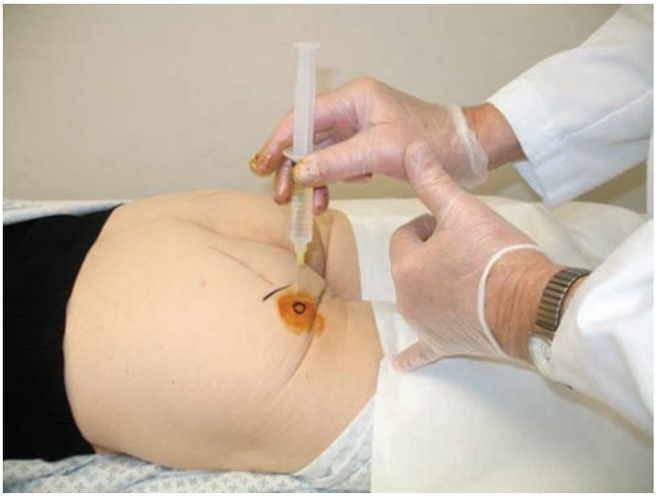

Figure 33 **Injection of the perineal branch of the left posterior femoral cutaneous nerve. After diagnosis, the site is marked. Infiltration of a bupivacaine 0.25% and lidocaine 1.0% mixture, 8 mL, may be augmented with steroids. Her persistent post-decompression pains in the transverse gluteal fold, near the anus and labia, and in the groin were relieved for "the first time in many years." This case (same patient as in Figure 31) demonstrates the need to pursue postoperative treatment of all neuropathies in the perimeter of the pudendal territory. (After decompression surgery, chronic constipation was relieved and her orgasms returned.)**

or, conversely, with clitorodynia. It is more common in females in our experience. Diagnosis is made by finger pressure 4 cm inferior to the ischial tuberosity. The neuropathy often resolves using bupivacaine and steroid injections [*see Figure 33*]. Surgical excision can eliminate the pain.[69]

INFREQUENT PAINFUL PELVIC NEUROPATHIES

Genitofemoral neuropathy is challenging to diagnose because of the multiple nerves affecting its "territory." It becomes a diagnosis by exclusion, requiring a nerve block to ascertain its causal role in the patient's pelvic pain.[70]

Obturator neuropathy, formerly infrequent, has become common because of gynecologic surgeries that use mesh placement via the obturator foramen. Immediate postoperative pain occurs in the medial thigh to the knee. Diagnosis is clinical, and transvaginal pressure over the obturator region often reproduces the pain. Surgical release is required and may provide immediate relief.[71]

Inferior cluneal neuropathy affects the lower buttock region. It is challenging to delineate this from the other neuropathies affecting that area, for example, MCN, posterior femoral neuropathy, and pudendal neuropathy. Treatments include infiltrations of local anesthetics or surgery.[72]

Superior cluneal neuropathy is also a cause of low back and buttock pain.

Portions of the genitalia are also innervated by the ilioinguinal and genitofemoral nerves. The perineum may have fibers from the perineal branch of the posterior femoral cutaneous nerve, the long perineal nerve, and others.

Financial Disclosures: Stanley J. Antolak Jr, MD, has no relevant financial relationships to disclose.

REFERENCE KEY

Review Clinical Trial Meta-analysis Guideline

References

1. Pecina MM, Krmpotic-Nemanic J, Markiewitz AD. Tunnel syndromes: peripheral nerve compression syndromes. 3rd ed. Boca Raton (FL): Taylor and Francis; 2001.
2. Hilton J. Rest and pain. London: G. Bell and Sons; 1918.
3. Robert R, Prat-pradat D, Labat JJ, et al. Anatomic basis of chronic perineal pain: role of the pudendal nerve. Surg Radiol Anat 1998;20:93–8.
4. Treede RD, Jensen TS, Campbell JN, et al. Redefinition of neuropathic pain and a grading system for clinical use: consensus statement on clinical and research diagnostic criteria. Neurology 2008;70:1630–5.
5. Wenninger K, Heiman JR, Rothman I, et al. Sickness impact of chronic nonbacterial prostatitis and its correlates. J Urol 1996;155:965–8.
6. ACOG Committee on Practice Bulletins—Gynecology. ACOG Practice Bulletin No. 51. Chronic pelvic pain. Obstet Gynecol 2004;103:589–605.
7. Litwin MS, Mcnaughton-Collins M, Fowler FJ Jr, et al. The National Institutes of Health Chronic Prostatitis Symptom Index: development and validation of a new outcomes measure. J Urol 1999;162:369–75.
8. Engeler D, Baranowski AP, Elneil S, et al. EAU guidelines. Presented at: 27th European Association of Urology Annual Congress; 2012 Feb 24–28, 2012; Paris, France.
9. Abrams P, Cardozo L, Fall M, Griffiths D, Rosier P, Ulmsted U, Van Kerrebroek P, Victor A, Wein A. The Standardization of Terminology in Lower Urinary Tract Function: Report of the Standardization Subcommittee of the International Continence Society. Urology 2003; 61: 37-49.
10. Hibner M, Castellanos ME, Desai N, et al. Glob Libr Womens Med 2011; (ISSN:1776-2228) DOI: 10.3843/GLOWM.10468.
11. Beco J, Climov D, Bex M. Pudendal nerve decompression in perineology; a case series. BMC Surg 2004;4:15.
12. Zhang R, Sutcliffe S, Giovanucci E, et al. Lifestyle and risk of chronic prostatitis/chronic pelvic pain syndrome in a cohort of United States male health professionals. J Urol 2015;194:1295–300.
13. Labat JJ, Robert R, Bensignor M, et al. [Neuralgia of the pudendal nerve. Anatomo-clinical considerations and therapeutical approach]. J Urol (Paris) 1990;96:239.
14. Clemens JQ. Afferent neurourology: an epidemiological perspective. J Urol 2010;18:432–9.
15. Farmer MA, Huang L, Martucci K, et al. Brain white matter abnormalities in female interstitial cystitis/bladder pain syndrome: a MAPP Network neuroimaging study. J Urol 2015;194:118–26.
16. Antolak S, Hough D, Pawlina W, Spinner R. Anatomical basis of chronic pelvic pain syndrome: the ischial spine and pudendal nerve entrapment. Med Hypotheses 2002;59:349–53.
17. Ukrainets IV, Mospanova EV, Davidenko AA, Shishkina SV. 4-Hydro-2-quinolones. 192.* Relationship of xy structure and analgesic activity of 4-amino-2-oxo-1,2-dihydroquinoline-3-

carboxylic acids and their derivatives. Chemistry of Heterocyclic Compounds 2011;46:1371–9.
18. Chen S, Hui H, Zhang D, Xue Y. The combination of morphine and minocycline may be a good treatment for intractable post-herpetic neuralgia. Med Hypotheses 2010;75:663–5.
19. Yilmaz U, Liu YW, Berger RE, Yang CC. Autonomic nervous system changes in men with chronic pelvic pain syndrome. J Urol 2007;177:2170–4.
20. Williams DP, Chelimsky G, McCabe NP, et al. Effects of chronic pelvic pain on heart rate variability in women. J Urol 2015;194:1289–94.
21. Zuelzer G. Reizung des Nervus pudendus (Neuralgie). Berlin Klin Woch 1915;49:1280–1.
22. Turner MLC, Marinoff SC. Pudendal neuralgia. Am J Obstet Gynecol 1991;165:1233–5.
23. Jacobs BO. Cutaneous pinprick sensibility as a screening device. Part Two: Enhanced diagnosis of diabetic peripheral neuropathy using refined technique and dedicated single use precision technology. Diabetic Microvascular Complications Today 2006;(July/August):33–36.
24. Antolak SJ, Antolak CM, Lendway L. Measuring the quality of pudendal nerve perineural injections. Pain Physician 2016;19:299–306.
25. Labat JJ, Riant T, Robert R, et al. Diagnostic criteria for pudendal neuralgia by pudendal nerve entrapment (Nantes criteria). Neurourol Urodyn 2008;27:306–10.
26. Antolak SJ, Antolak CM. Failed sacral neuromodulation: simple tests demonstrate pudendal neuropathy. A case series. J Pelvic Med Surg 2006;12:35–40.
27. Backonja M-M, Edwards R, Seghal N, et al. Quantitative sensory testing in measurement of neuropathic pain phenomena and other sensory abnormalities. Clin J Pain 2009;25:641–7.
28. Bleustein CB, Eckholdt E, Arezzo JC, Melman A. Quantitative somatosensory testing of the penis: optimizing the clinical neurological examination. J Urol 2003;169:2266–9.
29. Khalili N, Wendelschafer-Crabb G, Kennedy WR, Simone DA. Influence of thermode size for detecting heat pain dysfunction in a capsaicin model of epidermal nerve fiber loss. Pain 2001;91:241–50.
30. Vardi Y, Greunwald I, Sprecher E, et al. Normative values for female genital sensation. Urology 2000;56:1035–40.
31. Dyck PJ, O'Brien PC, Kosanke JL, et al. A 4, 2, and 1 stepping algorithm for quick and accurate estimation of cutaneous sensation threshold. Neurology 1993;43:1508–12.
32. Beco J, Seidel L, Albert A. Normative values of skin temperature and thermal sensory thresholds in the pudendal nerve territory. Neurourol Urodyn 2015;34:571–7.
33. Krumova EK, Geber C, Westermann A, Maier C. Neuropathic pain: is quantitative sensory testing helpful? Curr Diab Rep 2012;12:393–402.
34. Laurberg S, Swash M, Snooks SJ, Henry MM. Neurologic cause of idiopathic incontinence. Arch Neurol 1988;45:1250–3.
35. Tetzschner T, Sorensen M, Lose G, Christiansen J. Pudendal nerve function during pregnancy and after delivery. Int Urogynecol J Pelvic Floor Dysfunct 1997;8:66–8.
36. Ricciardi R, Mellgren AF, Madoff RD, et al. The utility of pudendal nerve terminal motor latencies in idiopathic incontinence. Dis Colon Rectum 2006;49:852–7.
37. Parnell BA, Howard JF Jr, Geller EJ. The effect of sacral neuromodulation on pudendal nerve function and female sexual function. Neurourol Urodyn 2015;34:456–60.
38. Shafik A, El-Sabai O, Shafik, A, Shafik IA. Role of pudendal canal syndrome in pathogenesis of interstitial cystitis and its treatment by pudendal canal decompression. Curr Urol 2008;2:24-29.
39. Bautrant E, de Bisschop E, Vaini-Elies V, et al. Modern algorithms for treating pudendal neuralgia; 212 cases and 104 decompressions. J Gynecol Obstet Biol Reprod (Paris) 2003; 32:705–12.
40. Benson JT. Neurophysiology of the female pelvic floor. Curr Opin Obstet Gynecol 1994;6:320–3.
41. Grigorescu BA, Lazarou G, Olson TR, et al. Innervation of the levator ani muscles: description of the nerve branches to the pubococcygeus, iliococcygeus, and puborectalis muscles. Int Urogynecol J Pelvic Floor Dysfunct 2008;19:107–16.
42. Hodges PW, McLean L, Hodder J. Insight into the function of the obturator internus muscle in humans: observations with development and validation of an electromyography recording technique. J Electromyogr Kinesiol 2014;24:489–96.
43. de Bisschop E, Bautraunt E. Exploration electrophysiologique perineale dans le cadre de la nevrale pudendale: nouveaux concepts. In: Proceedings of the Annual Meeting le Choix des Armes, Marseille, France, 10–11 March 2006. p. 1–8.
44. Mahahhanukrauh P, Surin P, Vaidhayakarn P. Anatomical study of the pudendal nerve adjacent to the sacrospinous ligament. Clin Anat 2005;18:200–5.
45. Nakanishi T. [Studies on the pudendal nerve. I. A macroscopic observation of the pudendal nerve in man]. Kaibogaku Zasshi (Acta Anat Jap) 1967;42:223–9.
46. Antolak SJ. Interventions in managing male pelvic pain. In: Manchikanti L, Singh V, editors. Interventional techniques in chronic non-spinal pain. Paducah (KY): ASIPP Publishing; 2009. p. 303–18.
47. Bensignor MF, Labat JJ, Robert R, Ducrot P. Diagnostic and therapeutic nerve blocks for patients with perineal non-malignant pain [abstract]. In: 8th World Congress on Pain. 1996. p. 56.
48. Kim SH, Song SG, Paek OJ, et al. Nerve-stimulator-guided pudendal nerve block by pararectal approach. Colorectal Dis 2012;14:611–5.
49. Roberts WH, Taylor WH. Inferior rectal nerve variations as it relates to pudendal block. Anat Rec 1973; 177:-461-463
50. Martínez LC, Casal PP, Prieto LA, et al. Pulsed radiofrequency on terminal branches of the pudendal nerve: preliminary results. J Anesth Clin Res 2015;6:523.
51. Shafik A. Pudendal canal syndrome: a new etiological factor in prostadynia and its treatment by pudendal canal [de]compression. Pain Digest 1998;8:32–6.
52. Erdogru T, Avci E, Akand M. Laparoscopic pudendal nerve decompression and transposition combined with omental flap protection of the nerve (Istanbul technique): technical description and feasibility analysis. Surg Endosc 2014;28:925–32.
53. 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;47:403–8.
54. Shafik A. The posterior approach in the treatment of pudendal canal syndrome. Coloproctology 1992;14:310–5.
55. Benson JT, McClellan E. The effect of vaginal dissection on the pudendal nerve. Obstet Gynecol 1993;387–9.
56. Barry MJ, Fowler FJ Jr, O'Leary MP, et al. The American Urological Association symptom index for benign prostatic hyperplasia. J Urol 1992;148:1549–57.

57. Rosen RC, Cappelleri JC, Smith MD, et al. Development and evaluation of an abridged, 5-item version of the International Index of Erectile Function (IIEF-5) as a diagnostic tool for erectile dysfunction. Int J Impot Res 1999;11:319–26.
58. Rosen RC, Brown C, Helman J, et al. The Female Sexual Function Index (FSFI): a multidimensional self-report instrument for the assessment of female sexual function. J Sex Marital Ther 2000;26:191–208.
59. Haanpää M, Attal N, Backonja M, Baron R, Bennett M, Bouhassira D, Cruccu G, Hansson P, Haythornthwaite JA, Iannetti GD, Jensen TS, Kauppila T, Nurmikko TJ, Rice AS, Rowbotham M, Serra J, Sommer C, Smith BH, Treede RD. NeuPSIG guidelines on neuropathic pain assessment. Pain. 2011;152:14-27.
60. Sturgeon JA, Darnall BD, Kao MC, Mackey SC. Physical and psychological correlates of fatigue and physical function: a Collaborative Health Outcomes Information Registry (CHOIR) Study. J Pain 2015;16:291–8.
61. Roche B, Robert-Yap J, Skala K, Zuffrey G. Pudendal Nerve compression Syndrome. Societa Italiana di Chirugia ColoRettale www.siccr.org 2009; 20:172-179.
62. Maigne R, Nieves W, editors. Diagnosis and treatment of pain of vertebral origin. 2nd ed. Boca Raton (FL): CRC Press; 2006.
63. Applegate WV. Abdominal cutaneous nerve entrapment syndrome. Surgery 1972;71:118–24.
64. Rab M, Ebmer J, Dellon AL. Anatomic variability of the ilioinguinal and genitofemoral nerve: implications for the treatment of groin pain. J Plast Reconstr Surg 2001;108:1618–23.
65. Hebbard P, Fujiwara Y, Shibata Y, Royse C. Ultrasound-guided transversus abdominis plane (TAP) block. Anaesth Intensive Care 2007;35:616–7.
66. Maigne JY, Maigne R. Trigger point of the posterior iliac crest: painful iliolumbar ligament insertion or cutaneous dorsal ramus pain? An anatomic study. Arch Phys Med Rehabil 1991;72:734–7.
67. Copeman WSC, Ackerman WL. Edema or herniations of fat lobules as a cause of lumbar and gluteal "fibrositis." Arch Intern Med 1947;79:22–35.
68. Ries E. Episacroiliac lipoma. Am J Obstet Gynecol 1937;34:492–8.
69. Tubbs RS, Miller J, Loukas M, et al. Surgical and anatomical landmarks for the perineal branch of the posterior femoral cutaneous nerve: implications in perineal pain syndromes. Laboratory investigation. J Neurosurg 2009;111:332–5.
70. Starling JR, Harms BA. Diagnosis and Treatment of Genitofemoral and Ilioinguinal Neuralgia. World J Surg 1989; 13:586-9
71. Tipton JS. Obturator neuropathy. Curr Rev Musculoskelet Med 2008;1:234–7.
72. Darnis B, Robert R, Labat JJ, et al. Perineal pain and inferior cluneal nerves: anatomy and surgery. Surg Radiol Anat 2008;30:177–83.

Acknowledgments

Figure 1a Courtesy of Dr. Kawanishi, Kagawa, Japan
Figure 17b and 17c Courtesy of Dr. Jeanette Potts.