



Pudendal Entrapment as an Etiology of Chronic Perineal Pain: Diagnosis and Treatment

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Aims: This study was conducted to evaluate pudendal entrapment as an etiology of chronic pain, a diagnostic protocol for pudendal entrapment, and clinical response to surgical decompression. **Methods:** A case series of 58 consecutive patients with a diagnosis of pudendal entrapment, based on clinical factors, neurophysiologic studies, and response to pudendal nerve infiltrations, is described. All patients were refractory to other treatment modalities. Patients were assessed before and after surgical decompression: degree of pain was assessed by visual analog scale (VAS) score, percent global overall improvement, and improved function and quality of life before surgery and 12 months or longer after surgery. **Results:** The primary presenting feature was progressive, chronic, intractable neuropathic pain in the perineum (ano-rectal and/or urogenital) that worsened with sitting. Other symptoms included urinary hesitancy, frequency, urgency, constipation/painful bowel movements, and sexual dysfunction. After surgical decompression, 35 (60%) patients were classified as responders, based on one of the following three criteria: a greater than 50% reduction in VAS score, a greater than 50% improvement in global assessment of pain, or a greater than 50% improvement in function and quality of life. **Conclusions:** Pudendal entrapment can be a cause of chronic, disabling perineal pain in both men and women. Since symptomatic patients seek medical care from many different medical specialists, a reliable diagnostic protocol should be established. For patients refractory to conventional interventions, surgical decompression of the pudendal nerve can improve pain-related symptoms and disability. With ongoing work on this subject, which is a difficult disorder to accurately diagnose and treat, a better awareness of pudendal entrapment across specialties will emerge. *Neurourol. Urodynam.* © 2007 Wiley-Liss, Inc.

Key words: disabling neuropathic pain; neurophysiologic studies; pudendal entrapment; pudendal nerve infiltrations; responders; surgical decompression

INTRODUCTION

Pudendal nerve entrapment is a cause of chronic, disabling, intractable perineal pain in both male and female patients. Neuropathic pain—burning, tearing, stabbing lightning-like, electrical, sharp shooting, foreign body sensation—in the distribution of the pudendal nerve is characterized by worsening when sitting (but not on a toilet seat), reduction when standing, and absence upon awakening in the morning and progression throughout the day. While cycling, childbirth, prolonged sitting, trauma, and certain exercises have been implicated, the etiology of pudendal entrapment requires further study.^{1–3} Conservative treatment includes perineal hyperprotection aimed at preventing recurrent trauma to the nerve, pharmacologic neuromodulation, and physiotherapy.

The anatomical basis of pudendal nerve entrapment has been described in detail elsewhere.^{1,4,5} By way of background, and in brief, the pudendal nerve is a mixed nerve (motor ~20%, sensory ~50%, autonomic ~30%) with three branches: dorsal nerve of the penis/clitoris, perineal nerve, and inferior anal nerve, all derived from sacral S2–S4 roots (mainly S3) (Fig. 1). It supplies the anal and urethral sphincters and pelvic floor muscles, including bulbospongiosis, and provides anal, perineal, and genital sensitivity. Thus, pudendal nerve entrapment can result in unilateral or bilateral perineal, scrotal, testicular, and penile (female homologous sites: vulval, vaginal, clitoral) pain.

The S2–S4 nerve root leaves the pelvic cavity and enters the gluteal region, crossing over the ischial spine into the perineal region, where it divides into its two terminal branches—dorsal nerve of the penis (or clitoris) and the perineal nerve (Fig. 1).⁴ It is in this zone, at the ischial spine, where compression of the nerve is likely. The perineal nerve can be entrapped ventrally by the sacrospinous ligament and dorsally by the sacrotuberous ligament (Fig. 2).⁵ Entrapment can also occur at the falciform process of the sacrotuberous ligament where it can be entrapped (Fig. 2) by obturator fascia

in the pudendal canal (Alcock's canal) (Fig. 3)⁵; by the piriformis muscle (Fig. 4) (narrows sciatic notch and compresses the nerve against the posterior edge at the sacrospinous ligament); and directly at the ischial spine.⁵

There is an obvious parallel between pudendal nerve and focal nerve entrapments (e.g., median nerve at carpal tunnel, ulnar nerve at elbow). However, their clinical presentations are different due to anatomical differences, due to repeated compression throughout the day while sitting, and different fiber types (visceral, somatic, motor, sensory, autonomic). Thus, neurophysiologic testing can be helpful in diagnosis. In this regard, the typical Snooks and Swash neurophysiologic technique involves stimulation around the ischial spine and recording at the anal sphincter.⁶ A different technique was used in this study: we recorded at bulbospongiosis (skeletal muscle), a muscle that is innervated by the perineal branch of the pudendal nerve. In addition to neurophysiologic testing, pudendal nerve blocks are also necessary for diagnosis of pudendal nerve entrapment. Afferent fibers from the viscera and skin converge toward the same neurons in the spinal cord. In addition, the sympathetic system can be activated by its visceral afferents. This pain pathway of the pudendal nerve is assessed by perineal bupivacaine and steroid injections of the nerve at common entrapment sites.

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Abbreviations: cm, centimeter; CMAP, compound muscle action potential; CT, computed tomography; EMG, electromyogram; ml, milliliter; msec, millisecond; PNTML, pudendal nerve terminal motor latency; S, sacral; SI, sacroiliac; VAS, visual analog scale

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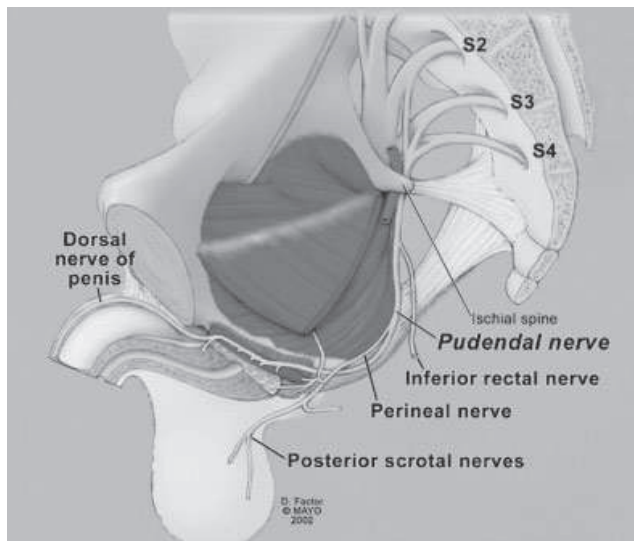


Fig. 1. Origin and branches of pudendal nerve.

MATERIALS AND METHODS

Study Participants

Study patients with pudendal nerve entrapment were identified using a diagnostic protocol that required clinical history (chronic, disabling, intractable neuropathic pain in the

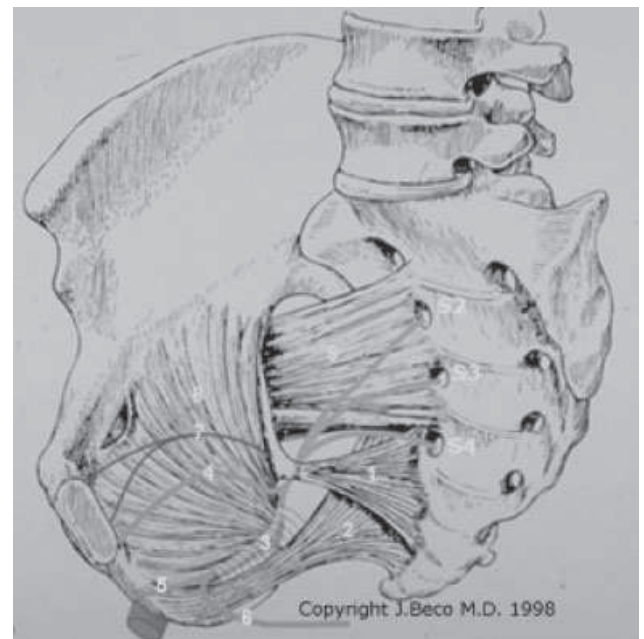


Fig. 3. (1) Sacro-spinal ligament; (2) Sacro-tuberous ligament; (3) Alcock's canal; (4) Nerve of the penis/clitoris; (5) Perineal branch of the pudendal nerve; (6) Anal branch of the pudendal nerve; (7) Arcus tendineus fascia pelvis; (8) Obturator muscle; (9) Piriformis muscle. S2, S3, and S4: Sacral roots forming the pudendal.

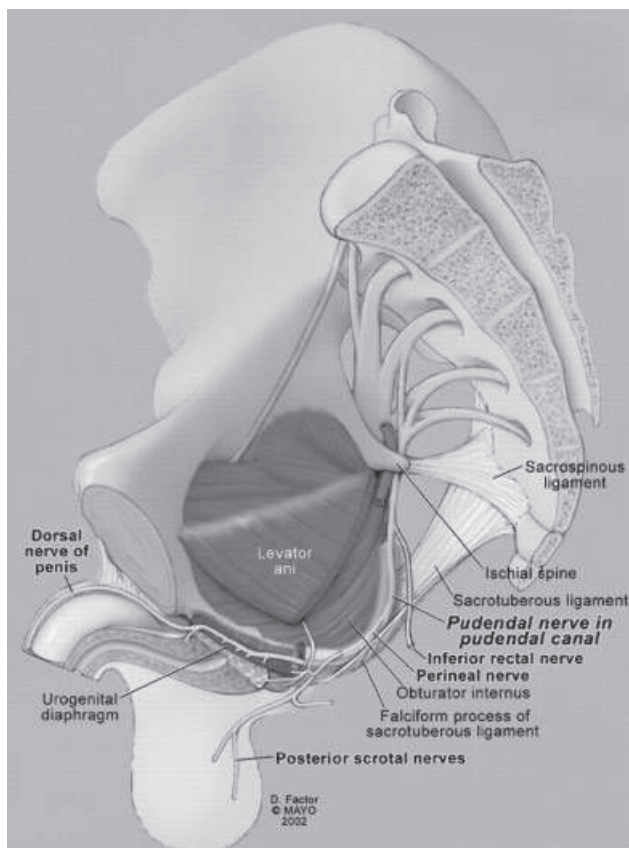


Fig. 2. Entrapment sites.

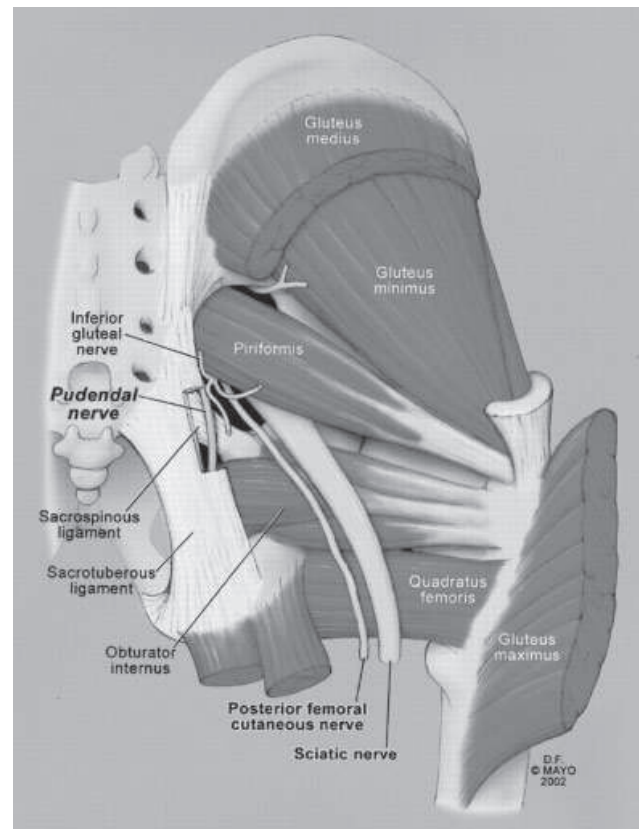


Fig. 4. Piriformis entrapment site.

distribution of the pudendal nerve aggravated by sitting) and either abnormal neurophysiology testing (prolonged pudendal nerve terminal motor latency [PNTML] or any EMG abnormality) or a response to the anesthetic portion of the pudendal block (A-delta and/or C-fibers or both) (Fig. 5).

Procedures and Measures

The protocol for diagnosis included three trials of computed tomography (CT)-guided pudendal nerve blocks (i.e., methylprednisolone [Depomedrol[®]] 40 mg followed by 5 ml of 1% lidocaine), separated from one another by 4 weeks and administered by the same physician.^{7,8} Each block was separated by 4 weeks. The blocks were administered bilaterally, the first two at the ischial spine and the third at Alcock's canal. Before and shortly after the diagnostic block, patients assessed their pain using a visual analog scale (VAS) of 0–10 for pain while sitting on a hard bench.

Neurophysiologic Testing. All patients had neurophysiologic testing done by the same physician (C.P.). Bilateral pudendal nerve distal motor latency tests (normal <4.0 mseconds (msec)⁹) and electromyogram (EMG) in pudendal-innervated muscles were performed. Acute and chronic denervation/reinnervation during EMG was recorded. Acute injury was defined as acute denervation with either increased insertional activity or fibrillations. Chronic injury was defined as chronic neurogenic change illustrated by chronic repetitive discharges, increased amplitude and long duration motor units, and polyphasia.

Surgical Decompression. Indications for surgery included a diagnosis of pudendal entrapment failed conservative treatment, and no lasting improvement from steroid pudendal nerve block (up to a few days for 80% of patients who provided response information) (Fig. 5).

The pudendal nerve was explored from its emergence from the pre-sacral region to its emergence from the Alcock's Canal. This was done via a trans-gluteal approach, using the method of Robert et al.¹⁰ and described as follows. A diagonal skin incision, measuring approximately 6–8 centimeters (cm), was centered 4 cm lateral to the sacrococcygeal junction. A muscle-splitting incision of the gluteus maximus muscle exposed the underlying sacrotuberous ligament, which forms a broad hood-like structure covering the underlying pudendal nerve and sacrospinous ligament. The upper margin of the ligament was identified, and a transverse incision was begun just lateral to the edge of the sacrum. The incision was carried caudad with care to identify and avoid injury to proximal branches of the nerve that are often found to course through the superficial layers of the sacrotuberous ligament.

When the pudendal nerve was located, the nerve branches were stimulated looking for compound muscle action potentials (CMAP) of the pudendal innervated muscles of the pelvic floor. Once the pudendal nerve was identified, a second transverse incision was made in the ligament lateral to where the nerve crosses the sacrospinous ligament. Sometimes the nerve, or a branch of it is found tethered to the underside of the ligament by fine fibrous tendrils that span from the surface of the nerve to the undersurface of the sacrotuberous ligament. Care must be taken to release the nerve without injury. The two transverse incisions of the sacrotuberous ligament were then continued onto the falciform process and then to the medial wall of Alcock's Canal, until there was no further fibrous tissue covering the nerve and its branches. The sacrospinous ligament was then

divided at its insertion on the ischial spine. (Care must be taken to ensure complete section of the sacrospinous ligament and associated vestigial muscle [just deep to the ligament]; otherwise a thin band might be left proximally, which can impinge the nerve as a knife-like process.) Finally, the pudendal nerve was freed of lateral attachments (usually one or two small branches of the pudendal artery and/or vein tether the neurovascular bundle laterally) and transposed anterior to the ischial spine. Care was taken not to injure the pudendal artery or vein. Transposition of the nerve is important, though, because the nerve often remains under some degree of tension until it is repositioned ventral to the ischial spine. The wound was then irrigated copiously with antibiotic solution and closed. The gluteus maximus was reapproximated with absorbable suture in its superficial fascia and the skin closed in layers.

Outcome Measures. At time of diagnosis, assessments of pain by VAS score and questions from the Impact Quality of Life (QOL) index of the NIH-CPSI¹¹ that related to function and quality of life were collected (Table I). These assessments were repeated at 12 months or longer following surgery at which time patients also completed a post-surgical questionnaire (Table I). The answers to these questions were converted to a numeric score to better quantitate results. In addition, patients were queried about surgical morbidity, improvement other than pain (urinary, sexual), and how long it took to resume most regular activities.

Statistical Analysis

The impact of potential prognostic factors (i.e., age, duration of symptoms and degree of prolonged distal latency) on surgical outcome (responder vs. non-responder) was evaluated using a two-sample, unpooled *t*-test. The impact of gender was evaluated by a *t*-test and confidence intervals and by Chi-square test for a 2 × 2 contingency table, with surgical success and gender as categories, testing for independence. Responders were defined as patients who met one of the following three criteria: had a 50% or greater reduction in VAS, a 50% or greater improvement in global assessment of pain, or a 50% or greater improvement in function and quality of life.

RESULTS

Fifty-eight consecutive patients (32 males and 26 females) were diagnosed with unilateral or bilateral pudendal entrapment. The mean (±SD) age was 46 (±11.8) years old. On average, patients were symptomatic for 3.9 (±3.9) years prior to treatment. All patients presented with a history of progressive, chronic, intractable neuropathic pain, which was located in the testicles, penis, or rectum in males and in the labia, clitoris, and rectum in females (Table II). Other baseline symptoms included urinary hesitancy, frequency, and urgency symptoms (40%); constipation (29%) including painful bowel movements; and sexual dysfunction (33%) (Table III). Most patients were severely disabled by their pain, with symptoms affecting quality of life and limiting patients' ability to engage in normal daily activities (Table IV preop data): 86% responded that they would be unhappy or feel terrible if "you were to spend the rest of your life with your symptoms just the way you have been" and 72% responded that their symptoms "kept you from doing the kind of things you would usually do over the last month". None of the patients had evidence of organ disease. Evaluations may have

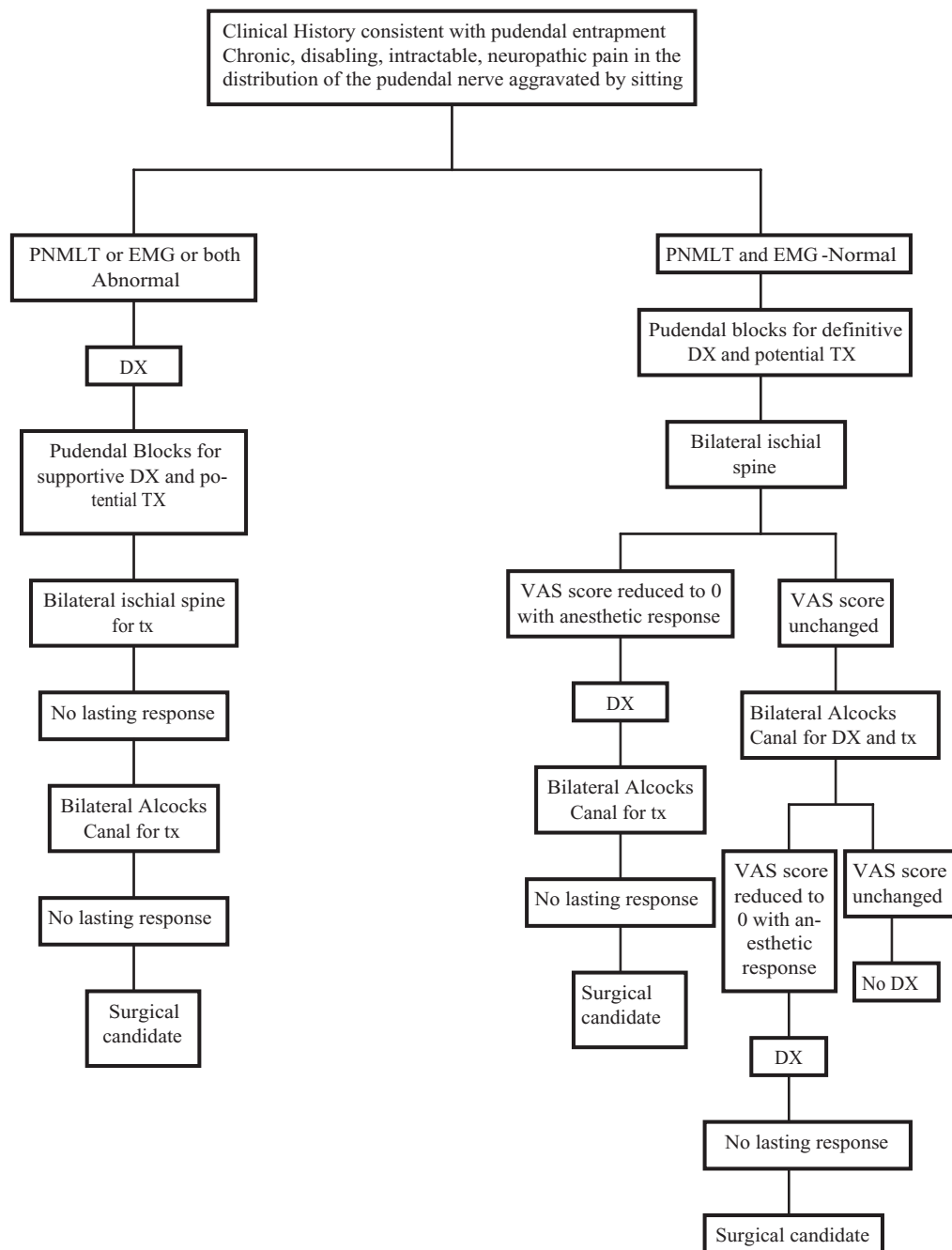


Fig. 5. Study procedures.

included negative work-up for prostatitis and epididymitis, negative scans of pelvis and lumbosacral spine, and normal findings on colorectal evaluation/laparoscopy.

Patients presented with the following diagnoses: interstitial cystitis (30%), prostatitis or epididymitis (63% of males), vulvadynia (50% of females), endometriosis (13% of females), piriformis syndrome (20%), levator ani syndrome (3%), coccydynia (6%), lumbosacral radiculopathy (3%), and chronic pelvic pain syndrome (20%). Mean pudendal nerve distal motor latency was 3.3 (± 1.7) msec, with 43% of patients having abnormal values (Table III). All patients in this study

had failed conservative treatment. Patients had seen multiple physicians and had failed multiple pharmacologic treatments (mean = 1.6 agents) as well as physiotherapy before surgery. None achieved long-lasting relief from the steroid component of nerve blocks.

Based upon a comparison of pre-operative and 12-month post-operative responses to survey questions (Table I), disability was compared (Table IV) and patients were classified as responders (35 patients, 60%) or non-responders (23 patients, 40%) (Table V). No prognostic factor was identified that predicted response to surgery. Surgical morbidity included

TABLE I. Pre-Operative and Post-Operative Questionnaires

Pre-operative	
• How often have you had pain or discomfort in the area over the last month?	
(a) Never	Score = 4
(b) Rarely	Score = 3.3
(c) Sometimes	Score = 2.66
(d) Often	Score = 2
(e) Usually	Score = 1.3
(f) Always	Score = 0.66
• How much have your symptoms kept you from doing the kinds of things you over the last month?	
(a) None	Score = 4
(b) Only a little	Score = 3
(c) Some	Score = 2
(d) A lot	Score = 1
• If you were to spend the rest of your life with your symptoms just the way they have been, how would you feel?	
(a) Delighted	Score = 4
(b) Pleased	Score = 3.5
(c) Mostly satisfied	Score = 3
(d) Mixed	Score = 2.5
(e) Mostly dissatisfied	Score = 2
(f) Unhappy	Score = 1.5
(g) Terrible	Score = 1
• VAS (0–10)	
Post-operative	
• When was your surgery?	
• From month to month following surgery, has your pain been decreasing? If so, what is your overall percent improvement?	
• How often have you had pain or discomfort in any of the previously afflicted areas since surgery?	
(a) Never	Score = 4
(b) Rarely	Score = 3.3
(c) Sometimes	Score = 2.66
(d) Often	Score = 2
(e) Usually	Score = 1.3
(f) Always	Score = 0.66
• Which number best describes your average pain or discomfort on the days you had it since surgery?	
VAS 0-10	
• How much have your symptoms kept you from doing the kinds of things you would usually do, since surgery?	
(a) None	Score = 4
(b) Only a little	Score = 3
(c) Some	Score = 2
(d) A lot	Score = 1
• How much do you think about your symptoms since surgery?	
(a) None	Score = 4
(b) Only a little	Score = 3
(c) Some	Score = 2
(d) A lot	Score = 1
• If you were to spend the rest of your life with your symptoms just the way they have been since surgery, how would you feel?	
(a) Delighted	Score = 4
(b) Pleased	Score = 3.5
(c) Mostly satisfied	Score = 3
(d) Mixed	Score = 2.5
(e) Mostly dissatisfied	Score = 2
(f) Unhappy	Score = 1.5
(g) Terrible	Score = 1
• Are you still on daily medications for your pain? If so, which ones? Have you been able to decrease the dosages?	
• Do you have any bowel, bladder incontinence, or other problems?	
• Do you have any areas of numbness related to the surgery?	
• Have you been diagnosed with SI join, hip, or lower back problems as a result of surgery?	
• Besides pain improvement, have any other problems improved since surgery, I.D., sexual, urinary, etc.	
• How long did it take to get back to most of your regular activities?	

numbness in a small patch of the pudendal nerve (vaginal, rectal, perineum areas) (7 patients, 12%), sacroiliac joint dysfunction (5, 8.6%), and transient urinary incontinence (1.2%).

DISCUSSION

Chronic pelvic pain syndrome is a complex problem for multiple specialists to whom affected patients present for

healthcare. Pudendal entrapment should be considered among patients with neuropathic pain in the pudendal nerve distribution (male—penis, testicles, perineum, rectum; female—labia, clitoris, perineum, rectum) that worsens with sitting but not when sitting on the toilet seat. The pain may, or may not, be associated with bladder, sexual, or rectal dysfunction. At the bedside both positive and negative sensory symptoms can be assessed (male—glans, posterior scrotum, and perianal; female—clitoris, labia, and perianal). In

TABLE II. Location of Pain

Males (%)		Females (%)	
Perineum	84	Perineum	75
Testicles	68	Labia	38
Penis	29	Clitoris	33
Rectum	39	Rectum	71

addition, application of pressure on the nerve at the ischial spine or Alcock's canal creates significant pain, supporting the diagnosis (Tinel's sign). Finally, autonomic dysfunction of the pudendal nerve can result in Sudomotor changes in gluteal skin (cutis anserina, peau d'orange) or Vasomotor changes (retracted penis).

Neurophysiologic studies and temporary, but not sustainable, pain relief with pudendal blocks help support the diagnosis of pudendal entrapment. The neurophysiologic tests conducted in this study were pudendal distal motor latency and EMG. With recording at bulbospongiosis, prolonged PNTML from chronic constipation may be eliminated. The PNTML is limited to detecting entrapment of fast conducting motor fibers between the stimulus and recording muscle (either bulbospongiosis and/or external anal sphincter). Additional testing using quantitative sensory testing and bulbocavernosus reflex (sacral reflex) may also be useful in assessing the pudendal nerve. Quantitative sensory testing assesses small/large sensory fibers.^{12,13} The bulbocavernosus reflex is a polysynaptic reflex stimulating sensory afferents to S2–S4 nerve roots to the external anal sphincter. Increased latency would indicate proximal rather than peripheral lesions. Further studies from our clinic using these modalities are forthcoming.

TABLE III. Patient Demographics and Clinical Findings

Feature	Study group
Overall	(N = 58)
Sex	
Female (%)	26 (45)
Male (%)	32 (55)
Age (years)	
Mean ($\pm$ SD)	45.71 (11.843)
Range	21–78
Time with symptoms before surgery (months)	
Mean ($\pm$ SD)	47.38 ($\pm$ 47.553)
Range	1–180
Additional symptoms	
Urinary hesitance, frequency, and urgency (%)	40
Constipation (%)	29
Sexual dysfunction (%)	33
Prior diagnosis (%)	
Interstitial cystitis (%)	30
Vulvodynia (%)	50
Endometriosis (%)	13
Prostatitis or epididymitis (%)	63
Piriformis syndrome (%)	20
Levator ani syndrome (%)	3
Coccydynia (%)	6
Lumbosacral radiculopathy (%)	3
Chronic pelvic pain syndrome (%)	20
Pre-surgical pain	
Average VAS before surgery (range)	6.1 (2–10)
Motor latency distal pudendal nerve (msec) ^a	
Mean ($\pm$ SD)	3.26 (1.711, n = 116)
Range	0.90–9.80

^aRight and left latencies were measured for each patient.

Our results with pudendal block were similar to those of Amarenco et al.¹⁴ In addition, 16 of our patients had blocks in nearby areas (lumbosacrum, pelvic area [i.e., lumbosacral epidurals], genitofemoral blocks). None of these blocks helped their pain acutely, sub-acutely, or on a long-term basis. This likely negates any placebo effect as related to the pudendal blocks. If multiple blocks, perineal hyperprotection, pharmacologic neuromodulation, and physiotherapy fail (i.e., sacroileac joint dysfunction causing the attached sacrotuberous and sacrospinous ligaments compressing the pudendal nerve), surgical decompression is a viable treatment option (Fig. 5). We were unable to identify risk factors that predict the success or failure of surgery.

In almost all of our explorations, we encountered anatomic variations that accounted for direct entrapment and nerve compression or tethering of the nerve to the lateral pelvic wall such that pelvic floor motions causes impingement of the nerve against relatively rigid ligamentous structures. These included: (1) a branch came off proximal to the ischial spine and coursed through the sacrotuberous ligament (these branches are often entrapped where they penetrate the ligament inferiorly); (2) a branch of one of the sacral nerve roots perforated the sacrospinous ligament just medial to the ischial spine and joined the main trunk of the nerve in such a fashion that the nerve was tethered at this point; (3) the nerve branched at the level of the ischial spine with each branch entering a separate Alcock's canal such that the branches were encased in the fascial tubes; (4) the ligaments were hypertrophied, and the nerve was under obvious tension when exposed; (5) the nerve was tethered laterally and dorsally by fine filaments bridging the nerve surface to the underside of the sacrotuberous ligament; and (6) there was diffuse thickening of the fascial planes with encasement of the nerve by the falciform process and Alcock's canal. Other areas that should be considered are entrapment proximal and distal to the common entrapment sites such as through piriformis muscle and at the urogenital diaphragm.¹⁵

The presumed pathology of pudendal entrapment could involve focal nerve damage (large fast conducting or small fibers) in the multiple areas of potential compression. Such a lesion can be maintained indefinitely if the compression is repeatedly renewed by continuous neural trauma from sitting. The varied presentation of pudendal entrapment reflects the nerve being mixed (motor, sensory, autonomic) and having multiple branches with anatomic variations. Patients' pain history typically reveals a remitting relapsing course that evolves into a chronic, progressive course. This may simply represent central sensitization being maintained by continued nerve compression, leading to chronic central sensitization.

CONCLUSION

Chronic, intractable, disabling perineal pain in men and women may be caused by pudendal entrapment. The diagnosis of this syndrome is not well established. We described a protocol for the diagnosis and treatment of pudendal entrapment, the first of its kind in the US, with a larger study from our clinic forthcoming. For patients refractory to conservative interventions, surgical decompression via a transgluteal approach can improve symptoms and disability. A continued look at non-responders who continue to experience pain after surgical decompression should be a priority of future research. The hypothesis of central sensitization continuing on after decompression is viable. With this in mind, botulinum toxin (Botox[®]) injections in the area of the

TABLE IV. Pre-Operative and Post-Operatives Results for Questionnaire

Pre-operative		Pre-op	Post-op
• How often have you had pain or discomfort in the area over the last month?		Mean: 1.0172	Mean: 1.6379
a. Never	(score = 4)	±SD: .5127	±SD: .7856
b. Rarely	(score = 3.3)		Difference: .6207
c. Sometimes	(score = 2.66)	Sum: 59	Sum: 95
d. Often	(score = 2)		%change: 61.0169
e. Usually	(score = 1.3)		
f. Always	(score = .66)		
• How much have your symptoms kept you from doing the kinds of things you over the last month?		Pre-op	Post-op
a. None	(score = 4)	Mean: 1.3621	Mean: 1.9655
b. Only a little	(score = 3)	±SD: .6407	±SD: 1.0081
c. Some	(score = 2)		Difference: .6034
d. A lot	(score = 1)	Sum: 79	Sum: 114
			%change: 44.3038
• If you were to spend the rest of your life with your symptoms just the way they have been, how would you feel?		Pre-op	Post-op
a. Delighted	(score = 4)	Mean: 1.2845	Mean: 2.0776
b. Pleased	(score = 3.5)	±SD: .4278	±SD: .9071
c. Mostly satisfied	(score = 3)		Difference: .7931
d. Mixed	(score = 2.5)	Sum: 74.5	Sum: 120.5
e. Mostly dissatisfied	(score = 2)		%change: 61.645
f. Unhappy	(score = 1.5)		
g. Terrible	(score = 1)		

TABLE V. Results for Responder Versus Non-Responders

Responders	(N = 35, 60%)
Sex	
Female (%)	14 (40)
Male (%)	21 (60)
Age (years)	
Mean (± SD)	45.77 (12.696)
Range	21–78
Time with symptoms before surgery (months)	
Mean (± SD)	43.49 (48.598)
Range	1–180
Motor latency distal pudendal nerve (msec)	
Mean (± SD)	3.28 (1.788, n = 70)
Range	0.90–9.80
Non-responders	(N = 23, 40%)
Sex	
Female (%)	12 (52)
Male (%)	11 (48)
Age (years)	
Mean (± SD)	45.61 (10.689)
Range	25–62
Time with symptoms before surgery (months)	
Mean (± SD)	53.30 (46.345)
Range	8–156
Motor latency distal pudendal nerve (msec) ^a	
Mean (± SD)	3.23 (1.606, n = 46)
Range	1–8.30

^aRight and left latencies were measured for each patient.

nerve could then stop central sensitization after decompression.¹⁶ This should be another area of study. With continued commitment to research, our understanding of and treatment for this painful, disabling pudendal entrapment will be refined.

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