Review Article

Pudendal Neuralgia

Michael Hibner, MD, PhD*, Nita Desai, MD, Loretta J. Robertson, PT, and May Nour, MD, PhD

From the Department of Obstetrics and Gynecology, St. Joseph's Hospital and Medical Center (all authors), Phoenix, Arizona.

ABSTRACT Pudendal neuralgia is a painful, neuropathic condition involving the dermatome of the pudendal nerve. This condition is not widely known and often unrecognized by many practitioners. The International Pudendal Neuropathy Association (ipna.org) estimates the incidence of this condition to be 1/100,000; however, most practitioners treating patients with this condition feel the actual rate of incidence may be significantly higher. Currently, there is a paucity of medical literature and scientific evidence in the diagnosis and treatment of pudendal neuralgia. Diagnosis of this condition is based on the utilization of Nantes Criteria, in conjunction with clinical history and physical findings. CT-scan guided nerve blocks are also employed by this author, to provide additional information. Subsequent treatment of pudendal neuralgia is medical and well as surgical, with Physical Therapy a key component to all aspects of treatment. The goal of this paper is to present evidence based information, as well as personal clinical experience, in treating approximately 200 patients with pudendal neuralgia. Journal of Minimally Invasive Gynecology (2010) 17, 148-153 © 2010 AAGL. All rights reserved.

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Pudendal neuralgia is a painful, neuropathic condition involving the dermatome of the pudendal nerve [1]. This condition is not widely known and often unrecognized by many practitioners. The International Pudendal Neuropathy Association (ipna.org) estimates the incidence of this condition to be 1/100,000 of the general population. Spinosa et al [2] document the incidence at 1% in the general population, affecting women more than men. Orphanet (www.orpha.net—a European website providing information about orphan drugs and rare diseases) states pudendal neuralgia affects 4% of patients undergoing consultation for pain and affects 7 women for every 3 men. These mixed results exemplify the inability to derive the precise incidence of pudendal neuralgia.

Most practitioners treating patients with this condition believe the actual rate of prevalence may be significantly higher than stated in existing literature. Currently, there is a paucity of medical literature and scientific evidence in the diagnosis and treatment of pudendal neuralgia. A Medline search by these authors revealed only 58 published studies with the key words “pudendal neuralgia” or “pudendal nerve entrapment.” The goal of this article is to present evidence-based information, as well as personal clinical experience, in treating approximately 200 patients with pudendal neuralgia.

Anatomy of the Pudendal Nerve

Pudendal nerve entrapment has been described to manifest itself symptomatically in a number of debilitating manners; therefore a clear understanding of the nerve anatomy and distribution is essential in diagnosis. The pudendal nerve carries motor, sensory, and autonomic fibers; subsequently, both afferent and efferent pathways are affected by its injury [3]. As described in the literature, the distribution of the pudendal nerve, in the perineum, is mediated by 3 branches derived from the sacral roots S2-S4 [1,4-6]. These branches are the dorsal nerve of the penis or clitoris, the perineal nerve, and the inferior anal nerve. On the basis of this pattern of distribution, damage to the pudendal nerve can result in either unilateral or bilateral pain in the female vulva, vagina, or clitoris, or, correspondingly, the male scrotum, testes, or penis. Affereintly, it has also been suggested that pudendal nerve injury may be responsible for inducing an increase in the C-fiber pathway of the bladder, leading to urinary incontinence and contributing to the development of overactive bladder syndrome [7,8]. In most cases, neuropathic pain will present, in both sexes, as an unrelenting, refractory perineal pain that is worsened by sitting and is progressive throughout the day [9].

Neural cross-talk has been implicated as a critical player in the convergence of sensory pathways in the pelvis, which
coordinates bowel, bladder, and sexual function [10,11]. Dysfunction of the pudendal nerve, caused by entrapment or compression, is therefore not only suspect, in generating this chronic, debilitating pain but also likely to negatively alter the interaction between pelvic organs and the afferent, efferent, and autonomic signals, which mediate their proper function.

Symptoms of Pudendal Neuralgia

Symptomatically, pudendal neuralgia is defined as a burning neuropathic pain in the distribution of the pudendal nerve, as described above. In brief, the pain is localized to the vulva, vagina, clitoris, perineum, and rectum in females (Fig 1) and to the glans penis, scrotum excluding testicles, perineum, and rectum in males [1]. Although the entire distribution of the pudendal nerve can be affected either unilaterally or bilaterally, if the neuralgia is more distal, affecting only a particular branch of the pudendal nerve, the pain may then be restricted to the clitoris only, vulva/vagina only, or rectum only [12]. A small percentage of patients may have pain outside the area of innervation of the pudendal nerve in addition to classic pudendal nerve symptoms [12]. In those cases, it may present as vague, neuropathic pain in the area of the lower abdomen, posterior thigh, or even the lower back and can be attributed to muscle spasm [12]. Patients with pudendal neuralgia often have associated symptoms such as urinary frequency and urgency, symptoms mimicking interstitial cystitis, dyspareunia, and persistent sexual arousal [13]. The pain, as in other cases of neuropathic pain, is burning, tingling, and numbing in nature. Patients have significant hyperalgesia (increased sensitivity and significant pain to mild painful stimulus), allodynia (pain in response to nonpainful stimulus), and paresthesias (sensation of tingling, prickling, or numbness, commonly known as “pins and needles”) [14]. Typically, symptoms are present when patients are sitting down and are much less severe or may even be absent when lying down or standing [14]. Anecdotally, there is significantly less pain when sitting on a toilet seat versus a chair. This phenomenon is believed to be associated with pressure applied to the ischial tuberosities rather than to the pelvic floor muscles. Patients usually awaken in the morning with minimal or no symptoms; however, the pain will increase as the day progresses. Often, patients will report the sensation of having a foreign body in the vagina or feeling as though they are sitting on an object, such as a tennis ball. One of the most common complaints is a sensation of a “hot poker” in the vagina or the rectum.

Causes of Pudendal Neuralgia

Pudendal neuralgia can be caused by mechanical injury to the nerve, viral infection, or immunologic processes [15]. In the case of mechanical injury to the nerve, most practitioners will refer to the condition as pudendal nerve entrapment. This “entrapment” may be caused by pelvic floor muscle spasm (levator ani or obturator internus), pressure from surrounding ligaments (sacrospinous, sacrotuberous), or scar tissue from trauma or surgeries involving the surrounding areas [16]. In patients who have undergone surgery, entrapment may be caused by mesh or suture directly injuring the nerve [17,18].

Historically, the first documented group of patients with symptoms of pudendal neuralgia were competitive cyclists. French physiatrist Gerard Amarenco [19-21], in 1988, specified the condition as “cyclist syndrome” or “Alcock’s canal syndrome.” Soon thereafter, the discovery of the occurrence of pudendal neuralgia in noncyclist patients led to the idea of pelvic trauma as a cause of the pain. In women, the 3 most common causes of pudendal nerve entrapment are surgical injury, pelvic trauma, and childbirth [22]. In our practice most of our female patients present with pudendal neuralgia as a result of previous gynecologic surgery, particularly vaginal surgery for prolapse or incontinence. The second most common presentation, in female patients, is a history of previous pelvic trauma, such as heavy lifting, falls injuring the back or buttocks, as well as patients inserting foreign objects rectally [23]. The least common manifestation is pudendalnerve neuralgia caused by vaginal childbirth in females [22]. Pelvic trauma, on the other hand, is responsible for the greatest cohort of pudendal neuralgia cases in males [24].

Diagnosis of Pudendal Neuralgia

Diagnosis of pudendal neuralgia is difficult and often based on the exclusion of other conditions causing pain generated from the muscles of the pelvic floor. The differential diagnoses for pudendal neuralgia include the following: vulvodynia, pelvic floor tension myalgia, interstitial cystitis, and neuralgias of other pelvic nerves such as the obturator, genitofemoral, or ilioinguinal nerves. The most important element of the diagnostic process is the history, followed by the physical examination. A thorough history must elucidate the

Fig. 1. Sensory distribution of the pudendal nerve: (a) pudendal nerve (b) inferior cluneal nerve (c) obturator nerve (d) ilioinguinal and genitofemoral nerve.
onset of pain, in particular, the nature of symptoms, associated symptoms, and alleviating and aggravating factors. Thereafter, the physical examination may further illuminate the cause of the symptoms. It is first necessary to rule out any obvious or visible lesions in the vulvar, perineal, or rectal areas. Sensation to touch and pinprick, similar to vulvodynia examinations, is assessed next. Bimanual pelvic examination then follows, with attention to the pelvic floor muscles, in particular the levator and obturator muscles, as well as tenderness of the bladder and sacrospinous ligaments. In patients with pudendal neuralgia, maximum tenderness, or a trigger point, can be produced by applying pressure to the ischial spine, which serves anatomically as the insertion site for the sacrospinous ligament. Palpation of this area can reproduce pain and symptoms as a positive Tinel’s sign. To date, there are no imaging studies that diagnose pudendal neuralgia. Pudendal nerve motor terminal latency testing is neither useful nor predictable, given a high rate of intraobserver and interobserver variability. Additionally there are many factors such as vaginal delivery and previous surgery that can alter the results of nerve motor latency testing [25,26].

In our practice, we use a series of 3 computer tomography (CT)-guided blocks of the pudendal nerve, a protocol described by Dr. Roger Robert (personal communication), to aid in our diagnosis and possible treatment of this condition. CT is used to localize the area of the pudendal nerve, and injections are then performed unilaterally or bilaterally on the basis of patient symptoms. The initial injection is performed without sedation to allow for communication between the patient and interventionist radiologist, regarding the severity and location of the pain during the procedure. In our practice, we consider any degree of pain relief, for any duration of time, whether a consequence of local anesthetic or steroid, to be diagnostic of pudendal neuralgia. CT-guided injections are preferred over vaginal injections to ensure higher precision in diagnosis. Patients who do not respond with immediate pain relief to CT-guided block of the pudendal nerve are considered not to be affected by pudendal neuralgia.

In 2008 Labat et al [12] published the “Nantes Criteria” for the diagnosis of pudendal neuralgia. Those criteria are now widely accepted in many practices treating patients with pudendal neuralgia, including ours. Nantes criteria are grouped into 4 categories: essential criteria, complimentary criteria, exclusion criteria, and associated signs. To be diagnosed with pudendal neuralgia, a patient must exhibit all 5 inclusion criteria, without any symptoms of the exclusion criteria.

Nantes Criteria for Diagnosis Pudendal Neuralgia

Inclusion Criteria

- Pain in the area innervated by the pudendal nerve—extending from anus to clitoris (or penis)
- Pain more severe when sitting
- Pain does not awaken patients from sleep
- Pain with no objective sensory impairment
- Pain relieved by diagnostic pudendal block

Complementary Diagnostic Criteria

- Pain characteristics: burning, shooting, stabbing, numbing
- Allodynia or hyperesthesia
- Sensation of foreign body in the rectum or vagina (sympathalgia)
- Pain progressively worse throughout the day
- Pain predominantly unilateral
- Pain triggered by defecation
- Significant tenderness around ischial spine on vaginal or rectal examination
- Abnormal neurophysiology testing (puodenal nerve motor latency testing) in men and nulliparous women

Exclusion Criteria

- Pain located exclusively in the coccygeal, glutal, pubic, or hypogastric area (without pain in the area of distribution of pudendal nerve)
- Pruritus
- Pain exclusively paroxysmal
- Abnormality on the imaging test (magnetic resonance imaging, CT, and others), which can account for the pain

Associated Signs

- Buttock pain (area around ischial tuberosity) with sitting
- Referred sciatic pain
- Pain referred to the medial side of the thigh
- Suprapubic pain
- Urinary frequency or pain with full bladder
- Pain after orgasm/ejaculation
- Dyspareunia or pain after intercourse
- Erectile dysfunction
- Normal pudendal nerve motor latency

The development of these criteria is an important step toward providing uniform consensus for diagnosis and future treatment of pudendal neuralgia. Of note are the inclusion and exclusion criteria that focus on the areas of innervation of the pudendal nerve, as well as the associated symptoms that focus on areas and symptoms not innervated by the pudendal nerve. These criteria reiterate a strong need for comprehensive knowledge of the pelvic anatomy, meticulous history-taking, and an organized and perceptive physical examination.

Treatment of Pudendal Neuralgia

Much like in the treatment of other nerve compression syndromes, the initial treatment of pudendal neuralgia should always be conservative. Patients diagnosed with this syndrome are initially offered oral medications and physical therapy. In our practice we typically initiate pharmacotherapy with oral pregabalin 75 mg, twice a day, and titrate the
dosage, as tolerated, to a maximum daily dose of 600 mg/d. Patients are also offered muscle relaxants. Although systemic medications such as tizanidine and cyclobenzaprine HCl may alleviate the pain, we find local muscle relaxants to be vastly superior. We use 2 types of local muscle relaxants: rectal belladonna and opium suppositories and vaginal diazepam suppositories. Both can be used up to twice a day to aid in pain relief.

In terms of physical therapy, the patient needs to be treated by a therapist specially trained to work with pelvic floor muscle dysfunction. The therapist addresses muscle imbalances, spasm, restricted tissues, and other dysfunctions by focusing on palpation and manual techniques, posture, range of motion, and strength of the pelvic, back, and hips. Therapy is administered in the form of “hands-on” techniques, exercises, stretching, and education. Most of these patients have significant muscle spasm and subsequent muscle shortening throughout the pelvic girdle. Physical therapists use a variety of manual techniques to help release the muscle spasms and lengthen these muscles. These methods include myofascial release, soft tissue and connective tissue mobilization, and trigger point release. The pelvic floor muscles can only be fully examined and treated with either intravaginal or intrarectal approaches and to the level of patient tolerance. Therapists may also use different modalities such as biofeedback, ultrasonography, or electrical stimulation to assist with their treatments. All patients are given a home exercise regimen that includes relaxation and lifestyle modifications to continue the benefits gained during office sessions. Lastly, for patients in whom physical therapy cannot be performed secondary to severe muscle spasm, Botulinum toxin A may be injected directly into the muscles to aid in muscle relaxation and increase tolerance to physical therapy [27].

If there is no improvement in the level of pain, patients are then offered CT-guided injections of the nerve. As mentioned previously, the injections can serve both diagnostic and therapeutic roles. Usually, patients in our practice receive a series of 3 unilateral or bilateral CT-guided injection into the pudendal canal (Alcock’s canal). The nerve blocks consist of a cocktail of 0.5% bupivacaine 5 to 7 mL and triamcinolone 80 mg. Typically, patients are offered a series of 3 injections, 6 weeks apart. Steroid is included in the injection to provide secondary pain relief by reducing inflammation, which usually occurs within 2 weeks of the injection. Again, only the first injection serves a diagnostic purpose, the remaining 2 are performed to deliver steroid around the nerve, which can be therapeutic. Although pain relief is incredibly subjective, if a patient does not have sufficient pain relief, that is, at a level to which they can return to normal daily function, after the series of 3 injections, we then offer surgical decompression of the nerve.

Surgical Pudendal Nerve Decompression

Current literature describes 4 described approaches to decompression of an entrapped pudendal nerve. The most commonly performed procedure is the transgluteal technique as described by Roger Robert [128]. It is the preferred technique at our institution and will be described in detail. Other known techniques are transschiorectal [29], perianal [6], and laparoscopic procedures, as described by Dr. Alfredo Nieves from Chattanooga, Tennessee. The transgluteal technique has gained popularity because it allows for the greatest visualization of the pudendal nerve, offering the most precise method for decompression. In our practice we apply a modified technique of the Robert approach. Patients are placed in prone jackknife position. An incision is then created across the gluteal region over the area of the sacrotuberous ligament. Fibers of the gluteus muscle are then separated longitudinally and occasionally cut to reach the sacrotuberous ligament. Once identified, the sacrotuberous ligament is then transected at its narrowest point (Fig. 2). The pudendal nerve is most often identified immediately below the level of the sacrotuberous ligament, and, in some patients, may be attached to the anterior surface of the ligament. In our institution we often use a NIMS monitor (Nerve Integrity Monitoring System; Medtronic, Minneapolis, MN) to aid in identification the nerve. The pudendal nerve is then decompressed along its entire length, from the piriformis muscle to Alcock’s canal. The pudendal nerve, as seen in our patients, is most often found entrapped between the sacrospinous sacrotuberous ligaments, or the falciform process of sacrotuberous ligament. In cases in which pudendal neuralgia began after sacrospinous ligament suspension, or other mesh placement for prolapse, suture or mesh material is often found directly entrapping the nerve. Once the nerve is visualized to be free, in our practice, we place a segment of Neuragen (Origin Biomed, Inc., Halifax, Nova Scotia, Canada), a nerve-protecting tubing made of a collagen matrix, to prevent recracking of the nerve. We also saturate the nerve in a platelet-rich plasma matrix, which has been shown to promote nerve healing in other nerve surgeries [30]. This completes the decompression of the nerve. We will then place a pain

Fig. 2. Anatomy of the pudendal nerve, trans-gluteal approach: (a) transected sacrotuberous ligament (b) sacrospinous ligament (c) obturator muscle (d) piriformis muscle (e) ischiium with ischial spine marked by * (f) pudendal nerve and vessels entering Alcock’s canal.
pump catheter (On-Q pain buster pump; I-Flow Corporation, Lake Forest, CA) along the course of the nerve. We believe this direct blockage of the nerve, for a prolonged period of time, such as 10 to 20 days, with bupivacaine, helps reverse the phenomenon of central sensitization to pain within the spinal cord. This medication is delivered at a rate of 2 mL/h. Lastly, the sacrospinous ligament is repaired with a graft of cadaveric gracilis tendon. This modification is of great importance to the procedure because, we believe, it improves the stability of the sacroiliac joint. After surgery, patients remain hospitalized overnight and during that course in time are expected and encouraged to ambulate as much as tolerated. The pain pump is kept in place for 20 days, and physical therapy is initiated 6 weeks after surgery. According to data from France, after surgical decompression, approximately 40% of patients will be pain free, 30% will have some improvement in pain, and 30% will have no change in pain level [31]. There is a 1% risk that pain may worsen after surgical decompression. The pudendal nerve does not recover immediately after surgery, and first expected improvement in pain level often is noted to occur 4 months after surgery [28,31]. Maximum improvement in pain level is achieved 12 to 18 months after surgery. Surgical outcomes are relative to the condition and duration of the nerve injury before surgery [28,31]. Patients with symptoms of pudendal neuralgia for greater than 10 years are less likely to recover because of the long duration of damage of the nerve, especially when compared to their counterparts with pain of less than 10 years time [28,31].

Summary and Highlights

Suspect pudendal neuralgia in a patient with burning pain in the vulva, vagina, clitoris, perineum, or rectum.

Pain is always more severe with sitting and relieved or improved by standing.

Onset is usually immediately after vaginal surgery, pelvic trauma, or childbirth.

Offer help to the patient as soon as possible with the belief that earlier treatment may provide better outcomes.

Ask patient to minimize activities that worsen the pain.

Do not offer transvaginal injections, because precision is less than CT-guided injections. Transvaginal injections with high-potency steroid may preclude doing additional injections with radiologic guidance.

References

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