Successful Treatment of Penile Numbness and Erectile Dysfunction Resulting From Pudendal Nerve Entrapment

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Pudendal nerve decompression surgery has not been studied or reported for the treatment of penile numbness in the absence of pain. Herein, we report a case of a male patient with chronic numbness of the penis and erectile dysfunction in the absence of pelvic pain who was found to have pudendal nerve entrapment. This patient was treated with surgical decompression of the pudendal nerves that resulted in the return of genital sensation and erections. Thus, we propose that pudendal nerve entrapment may be considered as a cause of penile numbness and that pudendal nerve decompression surgery in these patients may be effective. UROLOGY 134: 228–231, 2019. © 2019 Elsevier Inc.

Penile numbness is an unusual presentation that is often associated with significant medical pathologies such as pelvic trauma, spinal cord injury, or diabetic neuropathy. However, numbness of the penis or genitals may also be associated with pudendal nerve entrapment.[1] Pudendal neuralgia is a debilitating condition that leads to neuropathic pain in the penis, scrotum, perineum, and anus, especially with sitting.[2] It may result from mechanical impingement of the pudendal nerve, typically as it courses along the sacrospinous ligament and through Alcock's canal. Studies have demonstrated that transgluteal pudendal decompression surgery is a successful treatment for patients with chronic pain due to pudendal nerve entrapment.[3] Nevertheless, surgical decompression has not been studied or reported for the treatment of penile numbness in the absence of pain. We report an unusual case of a gentleman who presented with chronic penile numbness with erectile dysfunction (ED) in the absence of pain who was found to have pudendal nerve entrapment. This patient was treated with bilateral pudendal decompression surgery and ultimately experienced a return of genital sensation and erections.

CASE REPORT

A 39-year-old Caucasian male from out of state presented for a second opinion regarding his chief complaint of penile numbness. Symptoms began approximately 10 years ago, when he developed a gradual onset of intermittent pain located within the rectum and perineum. Over the years, the pain resolved, but he then developed a constant numbness of his penis. He also complained of bilateral numbness of the scrotum, perineum, and anus. He had associated ED and was unable to maintain an erection for penetration. His symptoms were consistent with severe ED. The patient scored 4 points on the Sexual Health Inventory for Men questionnaire after answering all questions except question number 4. On review of systems, he reported urinary frequency, constipation, fecal incontinence, and sexual dissatisfaction. He denied any urinary incontinence, pelvic pain, or pain with sitting. Additional past medical history was significant for depression. He had a prior comprehensive urologic evaluation including a normal cystoscopy and prostate-specific antigen (PSA). He was treated with antibiotics for suspected prostatitis without relief. Sildenafil was prescribed without improvement in sexual function. In addition, he was evaluated by colorectal surgery and had an anal sphincteroplasty with minimal relief of anal symptoms. Lastly, he underwent a comprehensive neurologic evaluation with normal pudendal nerve motor terminal latency (PNMTL) test bilaterally.

On physical examination, he was found to have no sensation to pinprick in the distribution of the bilateral pudendal nerves including the perianal area, perineum, scrotum, penile shaft, and glans. Examination of the pelvic floor revealed normal tone without tenderness, and there was no tenderness at the bilateral ischial spines. Magnetic resonance imaging (MRI) of the pelvis with and without contrast showed no structural abnormalities. The prostate was normal in size and the bladder was unremarkable without any pelvic or groin lymphadenopathy. There was no mass

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effect along the course of the pudendal nerves and no abnormalities of the coccyx, sacrum or lumbar spine.

Due to the intractability of his numbness and given that symptoms were localized to the distribution of the pudendal nerve in the absence of other known etiologies, pudendal nerve entrapment was suspected. After informed consent, the patient was scheduled for transgluteal pudendal neurolysis and transposition. The patient chose to be operated on the left side first, with plan to operate on his right side if there was notable improvement.

SURGERY AND TREATMENT COURSE

Left transgluteal pudendal neurolysis and transposition was performed as follows: the patient was placed in prone jack-knife position, and a transgluteal incision was made to expose the left sacrotuberous ligament. The sacrotuberous ligament was opened with a Z-tenotomy technique. Using a surgical microscope and the Nerve Integrity Monitoring System, the pudendal nerve was identified and found to be compressed by the falciform process of the sacrotuberous ligament within the interligamentous space (surgical space between the sacrotuberous and sacrospinous ligaments). The nerve was exposed and adhesiolysis was performed. The sacrospinous ligament was then transected and the pudendal nerve was transposed anteriorly to alleviate compression. The nerve was then wrapped with a nerve protector. The sacrotuberous ligament was then reapproximated and the incision was closed. There were no postoperative complications.

At his 6-month follow-up, the patient reported some return of sensation to the left pelvic region such that he appreciated a noticeable difference compared to the right side, which remained completely numb. Therefore, he underwent right transgluteal pudendal neurolysis and transposition as previously described. Intraoperative findings revealed entrapment of the right pudendal nerve by the falciform process of the sacrotuberous ligaments within the interligamentous space as well as compression by fibrosis within Alcock's canal. There were no postoperative complications.

Ten months postoperatively, he reported sensation on the left, but continued numbness on the right. Examination noted improved sensation to pinprick bilaterally but persistent decreased sensation to light touch on the right. He also continued to complain of fecal incontinence and ED. Therefore, there was concern for persistent entrapment of the right pudendal nerve. The patient elected to pursue alternative management. He was evaluated by another specialist who placed a temporary InterStim (sacral neuromodulator) but reported no significant improvement in his numbness. He also underwent pelvic floor physical therapy. Two years from his initial surgery, he had a MR sacral plexus without IV contrast at an outside institution that showed mild enlargement of the right pudendal nerve with increased signaling suggesting possible entrapment. Thus, the patient underwent a repeat right transgluteal pudendal neurolysis. Intraoperative findings revealed severe dense adhesions at the entrance of Alcock's canal and within the interligamentous space (Fig. 1). The adhesions were lysed successfully. The patient had a normal postoperative course without complications and reported gradual improvement of his symptoms.

At a follow-up 2 years later, the patient reported he had regained satisfactory sensation of his penis as well as his perineum, scrotum, and anus. Although he was not administered a postoperative Sexual Health Inventory for Men questionnaire, he expressed renewed fulfillment and satisfaction with his sexual function and was able to maintain an erection for sexual intercourse. While he reported occasional use of tadalafil, he reported no use of medical therapy for ED at his

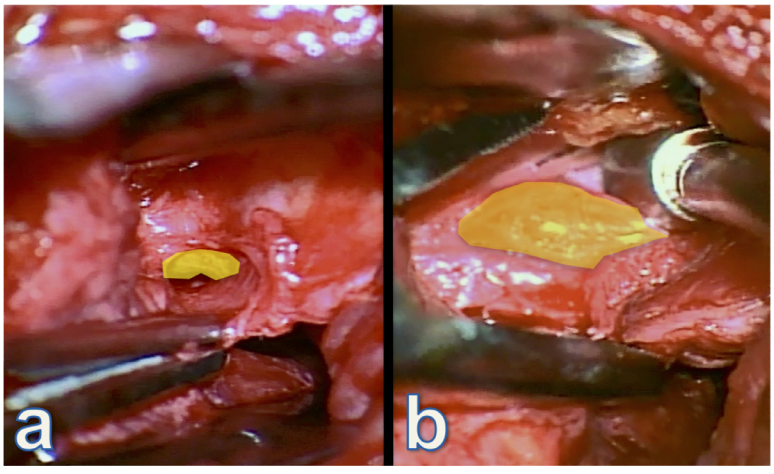

Figure 1. View of the interligamentous space using a surgical microscope after the sacrotuberous ligament was cut and reflected open. The pudendal nerve is highlighted in yellow. The right of the image is cephalad. The top of the image is medial. (**a**) The pudendal nerve identified within fibrotic tissue. (**b**) The pudendal nerve is exposed during neurolysis. (Color version available online).

3-year follow-up. Also, he reported resolution of fecal incontinence and urinary frequency.

DISCUSSION

The unique association between pudendal nerve impingement and penile numbness was first described in 1987 by Amarenco et al.[4] This landmark French study observed transient penile, scrotal, and perianal paresthesia in 5 cyclists, all of whom recently completed long bicycle tours. The authors proposed that the bicycle seat mechanically compresses the pudendal nerve as it courses ventromedially within Alcock's canal.

The pudendal nerve, originating from ventral spinal roots S2-4, carries motor, sensory, and autonomic fibers.[2,5] It courses from the greater sciatic foramen to pass through the interligamentous space across the sacrospinous ligament. At this point, it enters Alcock's canal, which is formed by the aponeurosis of the obturator internus muscle, where it usually divides into 3 branches: inferior rectal, perineal, and dorsal/penile.[5] The pudendal nerve controls the external anal sphincter, external urethral sphincter, and the muscles of the perineum and pelvic floor. It supplies sensation to the penis, perineum, scrotum, and rectum. Carrying both sympathetic and parasympathetic fibers, it suppresses bladder overactivity and is involved in the physiological changes during the sexual response.

Due to its narrow anatomic course, the pudendal nerve is uniquely prone to entrapment. The most common locations of entrapment are within the interligamentous space and within Alcock's canal.[6] Entrapment itself can result from several etiologies including cycling, sports, falls, trauma, pelvic reconstructive surgery, and vaginal deliveries. Interestingly, many patients are unable to identify an inciting event and report gradual onset of symptoms.

Classically, pudendal neuralgia is characterized by searing/burning/stabbing pains localized in the area of the penis/clitoris, scrotum/vagina, perineum, and rectum. It may be unilateral or bilateral. The hallmark symptom is neuropathic pain with sitting that is alleviated by standing. Associated symptoms included the sensation of a foreign body in the rectum, urinary frequency, ED, and numbness.[7]

Pudendal neuralgia is a clinical diagnosis that should be suspected in any patient presenting with pain and/or numbness in the dermatomal distribution of the pudendal nerve. To diagnose pudendal neuralgia, the clinician should look for the presence and absence of signs and symptoms as listed in Nantes Criteria (Table 1). Nantes Criteria are the clinical criteria that are regarded as the standard for the diagnosis of pudendal neuralgia by pudendal nerve entrapment.[1] The diagnosis of pudendal neuralgia can be confirmed by a pudendal nerve block that results in the relief of pain in the pudendal dermatome. Nevertheless, the block is nonspecific and does not predict successful treatment outcomes from decompression surgery.[8]

Table 1. Nantes criteria

Essential Criteria
Pain in the territory of the pudendal nerve: from the anus to the penis or clitoris
Pain is predominantly experienced while sitting
The pain does not wake the patient at night
Pain with no objective sensory impairment
Pain relieved by diagnostic pudendal nerve block
Complementary diagnostic criteria
Burning, shooting, stabbing pain, numbness
Allodynia or hyperpathia
Rectal or vaginal foreign body sensation (sympathalgia)
Worsening of pain during the day
Predominantly unilateral pain
Pain triggered by defecation
Presence of exquisite tenderness on palpation of the ischial spine
Clinical neurophysiology findings in men or nulliparous women
Exclusion criteria
Exclusively coccygeal, gluteal, pubic or hypogastric pain
Pruritus
Exclusively paroxysmal pain
Imaging abnormalities able to account for the pain
Associated signs not excluding the diagnosis
Buttock pain on sitting
Referred sciatic pain
Pain referred to the medial aspect of the thigh
Suprapubic pain
Urinary frequency and/or pain on a full bladder
Pain occurring after ejaculation
Dyspareunia and/or pain after sexual intercourse
Erectile dysfunction
Normal clinical neurophysiology

Nerve conduction studies, such as the PNMTL test, have been used previously for diagnosis, but findings are often unremarkable. Le Tallec de Certaines et al found no association between lengthening of the PNMTL and compressive symptoms in a cohort of 53 patients.[9] In addition, these nerve conduction tests were normal in the symptomatic cases of Amarenco et al.[4]

This patient's unique presentation posed a challenge for proper diagnosis and treatment in light of the current understanding of pudendal nerve compression and sensory deficits. Nantes Criteria state that patients should not have any objective sensory impairment since the dermatome of the pudendal nerve has sensory overlap with the genitofemoral, posterior femoral cutaneous and ilioinguinal nerves. Sensory impairment is usually suggestive of a sacral nerve root lesion, either within the conus medullaris or cauda equina. These lesions typically do not cause genital pain but lead to saddle anesthesia. Nevertheless, our patient did not demonstrate any evidence of cord compression or sacral nerve root impingement to explain his sensory deficits.

The key aspect in this patient's history that helped to confirm the diagnosis of pudendal neuralgia was his prior history of pain within the perineum and rectum. Thus,

pudendal nerve entrapment was suspected and transgluteal pudendal decompression surgery was offered. The transgluteal approach to pudendal neurolysis is the only surgical approach to be studied in a randomized clinical trial.[3] The laparoscopic approach, with or without robotic assistance, to pudendal decompression has also been described in small studies; but direct comparison studies are needed before this approach is recommended.[10,11] For pelvic pain, transgluteal pudendal neurolysis was effective in 71% of patients.[3] Surgery is designed to alleviate compression of the nerve thus allowing it to heal. The nerve may take up to 18 months to heal, with most patients having notable improvement 6 months postoperatively. There are no reported outcomes of pudendal decompression surgery for genital numbness in the absence of pain. Despite not meeting Nantes Criteria, our patient was found to have bilateral pudendal nerve entrapment and we demonstrated that decompression of his nerves resulted in return of sensation and sexual response.

After having a successful decompression of his left pudendal nerve, our patient did not have further clinical improvement after decompression of the right pudendal nerve. Proposed reasons for surgical failure include diagnostic error, compression causing irreversible nerve damage, incomplete release of the nerve, and/or postoperative fibrosis.[3] In this case, subsequent MR imaging identified signaling consistent with pudendal nerve entrapment from fibrosis. MR has been studied for its utility in the diagnosis of pudendal nerve entrapment, but findings may be inconsistent with surgical observation or surgical success.[12] Repeat surgery has been shown to be effective in select patients who have persistent symptoms after their initial surgery or who have a recurrence of symptoms.[13] Alternatives to surgery, such as pulsed radiofrequency ablation, which has demonstrated effectiveness for the treatment of pudendal neuralgia,[14] were not considered in this patient since he did not have any pain. Therefore, repeat surgery was offered, and it was found that the patient had developed extensive fibrosis leading to persistence of his symptoms.

Based on our experience with this patient, we propose that pudendal nerve entrapment may be a cause of penile numbness with ED in the absence of pain. Men with penile numbness, who are without diagnosis, may have unrecognized pudendal nerve entrapment and therefore should be evaluated for pudendal nerve injury. If pudendal nerve entrapment is suspected, pudendal decompression surgery may alleviate symptoms in these select patients.

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