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Repeat Operation for Treatment of Persistent Pudendal Nerve Entrapment After Pudendal Neurolysis

Michael Hibner, MD, PhD, Mario E. Castellanos, MD*, David Drachman, PhD, and James Balducci, MD, MBA

From the Department of Obstetrics and Gynecology (Drs. Hibner, Castellanos, and Balducci), St. Joseph's Hospital and Medical Center, Creighton University School of Medicine, Phoenix Campus, Phoenix, Arizona, and Maricopa Integrated Health System (Dr. Drachman), Phoenix, Arizona.

ABSTRACT **Study Objectives:** To describe a new approach to transgluteal pudendal neurolysis and transposition and to review the outcome in 10 patients who underwent repeat operation because of persistent pudendal neuralgia after failing to improve after initial surgical decompression.

Design: Retrospective analysis (Canadian Task Force classification II-3).

Setting: Academic chronic pelvic pain practice at St. Joseph's Hospital and Medical Center in Phoenix, Arizona.

Patients: Women and men with persistent pudendal neuralgia after undergoing transgluteal pudendal neurolysis and transposition.

Intervention: Transgluteal decompression of the pudendal nerve was performed in all 10 patients. In brief, a transgluteal incision was made, and the pudendal nerve was identified via a nerve integrity monitoring system. Adhesiolysis was performed from the piriformis muscle to the distal Alcock canal using a Zeiss NC-4 surgical microscope. The nerve was then enclosed in NeuraWrap Nerve Protector and coated with activated platelet-rich plasma. An ON-Q PainBuster catheter was place along the nerve into the Alcock canal, and 0.5% bupivacaine was infused at 2 mL/hr. The sacrotuberous ligament was repaired using an Achilles or gracillis cadaver ligament. The overlying subcutaneous tissue and skin were then closed.

Measurements and Main Results: From June 2008 to March 2010, 10 consecutive patients (7 women and 3 men; age range, 29–81 years) underwent repeat operation with transgluteal decompression of the pudendal nerve. Neuropathic pain was unilateral (n = 8) or bilateral (n = 2), in the clitoris or penis (30%), vulva or scrotum (70%), perineum (40%), and rectum (50%). Of the 10 patients, 1 patient was lost to follow-up. Mean follow-up was 23 months. Eight of 9 patients reported global improvement, with 2 patients reporting complete resolution of symptoms. One patient reported no change. Pain, as measured using an 11-point numerical scale, improved from a mean of 7.2 to 4.0 (p = .02), with 5 patients reporting clinically significant improvement (change, ≥2). Comfortable sitting or maximum time that the patient was able to sit without exacerbation of pain improved in 8 patients, with a change in median time of 5 to 45 minutes (p = .008). Change in the ability to sit correlated well with patient-reported global improvement (correlation coefficient, 0.86). No patient experienced worsening of symptoms.

Conclusion: Patients with persistent pudendal neuralgia after surgical decompression may benefit from repeat operation via our novel approach. Ability to sit correlates well with reported improvement due to surgery. Journal of Minimally Invasive Gynecology (2012) ■, ■–■

Keywords: Pudendal neuralgia; Pudendal nerve entrapment; Pelvic pain; Pudendal neurolysis; Platelet rich plasma

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Corresponding author: Mario E. Castellanos, MD, Department of Obstetrics and Gynecology, Arizona Center for Chronic Pelvic Pain, St. Joseph's Hospital and Medical Center, 500 W Thomas Rd, Ste 680, Phoenix, AZ 85013.

E-mail: Mario.castellanos@chw.edu

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Pudendal neuralgia is a severely painful and disabling neuropathic pain syndrome, affecting both women and men, in the distribution of the pudendal nerve. It is a rare disease, in which patients experience burning, tingling, or itching in the clitoris or penis, vulva or scrotum, perineum, or rectum. Symptoms may be unilateral or bilateral, and are classically exacerbated with sitting and alleviated with standing. The pudendal nerve is derived from the sacral root of S2–S4, and travels inferiorly and posteriorly in the fixed space between the sacrospinous and sacrotuberous ligaments. It then exits the pelvis through the obturator internus muscle membrane or Alcock canal. Along its course, the membrane divides into 3 branches, the dorsal clitoral/penile, perineal, and rectal, that carry motor, sensory, and autonomic fibers. Diagnosis of pudendal neuralgia is clinical, although block of the pudendal nerve can confirm the diagnosis by causing a transient resolution of symptoms. Initial treatments consist of behavior modification, analgesics, physical therapy, and additional nerve blocks. When these conservative treatments fail, surgical decompression is performed to treat pudendal nerve entrapment [1].

Pudendal neuralgia may develop from ligamentous entrapment between the sacrotuberous and sacrospinous ligaments [2]. In 2008, Labat et al [3] published a series of patients who benefited from transgluteal decompression and transposition of the pudendal nerve. They proposed the Nantes criteria (Table 1) for diagnosis, and reported that 70% of patients meeting these criteria improve with surgery. In a randomized controlled trial, 71% of patients had a positive outcome with transgluteal decompression of the pudendal nerve, compared with 13% who were managed conservatively [4]. Maximum improvement was observed at 12 to 18 months postoperatively, with 29% of patients reporting complete resolution of pain. Despite the high number of positive outcomes, about one-third of patients will not benefit from surgery, and as many as an additional one-third experience recurrence of pudendal neuralgia. Failure of surgery may be secondary to irreversible nerve damage from prolonged compression, incomplete release of the pudendal nerve, or postoperative fibrosis causing entrapment [4].

At our institution, we have made several modifications to address these surgical failures in the hope of improving outcomes. These modifications were designed to improve neurolysis, nerve regeneration, and adhesion prevention. Therefore, we offered repeat operation to patients in whom surgical decompression had previously failed. Herein, we describe our approach to pudendal neurolysis and review the clinical findings and outcomes of our first 10 consecutive patients who underwent repeat operation after failure of the initial surgical decompression procedure.

Materials and Methods

Through a retrospective review of medical records, we identified our first 10 consecutive patients who underwent repeat operation because of persistent pudendal neuralgia via transgluteal decompression of the pudendal nerve from June 2008 to March 2010. Because maximum improvement was observed at 12 to 18 months after surgery, patients operated on after March 2010 were excluded from the study because of less than 12 months of follow-up. A systematic review of the medical records, including office visit notes, operative reports, and follow-up telephone calls was performed. The present study was found exempt from review by our institutional review board because all information was obtained by review of medical records and patient identifiers were not recorded.

In all patients, transgluteal decompression of the pudendal nerve had failed, as previously described [4]. In brief, the procedure is performed by first making a gluteal incision from the sacrum to the ischial tuberosity, and is carried down to the sacrotuberous ligament. The ligament is then transected at this narrowest portion over the sacrospinous ligament. The pudendal neurovascular bundle is identified, separated from the posterior surface of the sacrospinous ligament, and digitally released throughout the course of the nerve. The sacrospinous ligament is then cut, and the nerve is transposed anteriorly. The subcutaneous tissue and skin are then closed.

Table 1

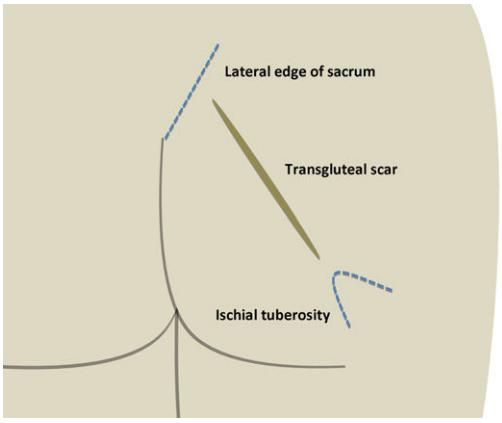
Nantes criteria for diagnosis of pudendal nerve entrapment

Inclusion criteria
Pain in area innervated by the pudendal nerve
Pain more severe with sitting
Pain does not awaken patient from sleep
Pain with no objective sensory impairment
Pain relieved by diagnostic pudendal block
Exclusion criteria
Pain located exclusively in coccygeal, gluteal, pubic, or hypogastric area (without pain in area of distribution of pudendal nerve)
Pruritus
Pain exclusively paroxysmal
Abnormality on imaging (CT, MRI) that can account for pain
Complementary criteria
Pain characteristics: burning, shooting, numbing
Allodynia or hyperesthesia
Allotriesthesia
Pain progressively throughout the day
Pain predominantly unilateral
Pain triggered by defecation
Significant tenderness around ischial spine
Abnormal neurophysiology test results
Associated signs/symptoms
Buttock pain (around ischial tuberosity)
Referred sciatic pain
Pain referred to medial side of thigh
Suprapubic pain
Urinary frequency with full bladder
Pain after orgasm/ejaculation
Dyspareunia or pain after intercourse
Erectile dysfunction

CT = computed tomography; MRI = magnetic resonance imaging.

Fig. 1

Buttocks with right transgluteal scar. Previous incision was made over the sacrotuberous ligament using the edge of the sacrum and ischial tuberosity as bony landmarks.



In all patients, the initial diagnosis was pudendal neuralgia, based on the Nantes criteria (Table 1). History and findings at physical examination in our office were consistent with continued pudendal neuralgia. Before undergoing repeat pudendal nerve decompression, most patients were treated conservatively using pudendal nerve blocks, medications, physical therapy, and Botulinum toxin A injections into pelvic floor muscles. Conservative management failed in all patients, and repeat operation was offered.

All transgluteal decompressions were unilateral, and were performed at St. Joseph's Hospital and Medical Center, Phoenix, Arizona. The patients were positioned in a prone jackknife position. The surface electrode of the nerve integrity monitoring system (NIM; Medtronic Xomed, Inc., Jacksonville, FL) was placed in the external anal sphincter, ipsilateral to the side of surgery, to monitor activity of the pudendal nerve. A transgluteal incision was made over the previous incision, and the gluteal muscles were separated down to the level of the sacrotuberous ligament (Fig. 1). In all patients, the ligament had been previously transected and not repaired. This interligamentous space (Fig. 2) was retracted and explored using the Zeiss NC-4 surgical microscope (Carl Zeiss Meditec, Inc., Dublin, California). The monitoring needle of the nerve integrity monitoring system was used to probe the interligamentous space (Fig. 3). The nerve was identified by registering activity at the external anal sphincter when touched. The nerve was isolated, and neurolysis was preformed using sharp dissection, releasing the nerve from adhesions from the inferior edge of the piriformis muscles to the distal Alcock canal (Fig. 2). A nerve-protector conduit (NeuraWrap Nerve Protector; Integra

Fig. 2

Open transgluteal incision reveals the interligamentous space. The pudendal neurovascular bundle is identified from the inferior edge of the piriformis muscle to the distal Alcock canal within the obturator internus muscle. Edges of previously severed ligaments are identified.

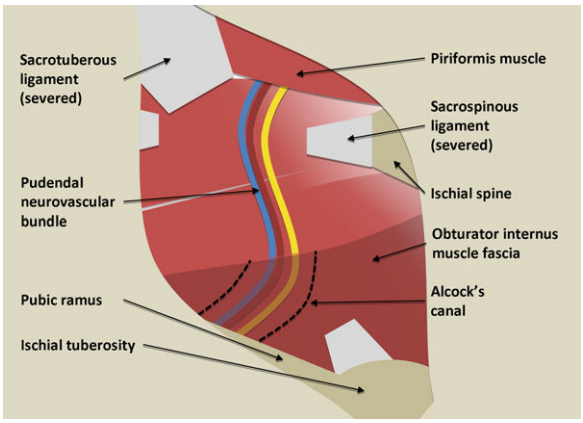


Fig. 3

The nerve integrity monitoring system (NIM) is used to identify the pudendal nerve. When the monitoring needle stimulates the pudendal nerve, activity is detected by the surface electrode inserted into the external anal sphincter muscle.

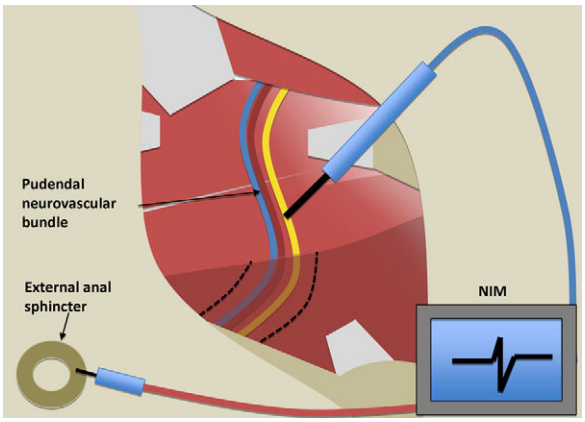


Fig. 4

After pudendal neurolysis, the nerve is enveloped with a NeuraWrap Nerve Protector, and activated platelet-rich plasma is injected around the nerve.

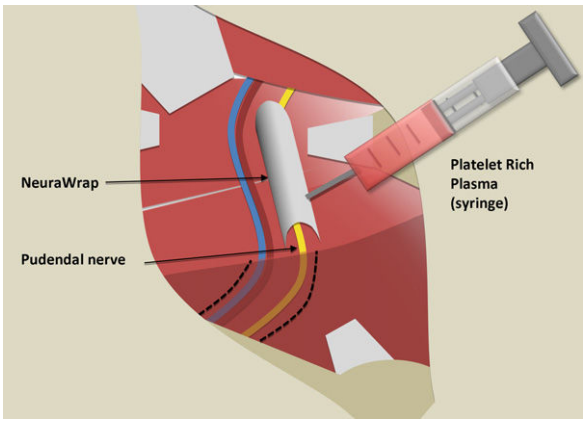
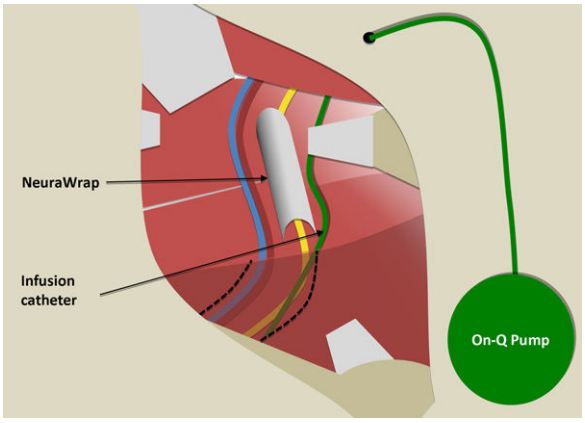


Fig. 5

The infusion catheter supplied with the ON-Q PainBuster is introduced above the gluteal incision and threaded along the wrapped nerve into the Alcock canal.



LifeSciences Holdings Corp., Plainsboro, NJ) was placed around the entire length of the exposed nerve to prevent adhesions. The nerve was then transposed anteriorly in the space previously occupied by the sacrospinous ligament to decrease tension. It was also coated with activated platelet-rich plasma, prepared from 120 mL of autologous blood drawn intraoperatively (Fig. 4). An infusion catheter (ON-Q PainBuster; I-Flow Corp., Lake Forest, CA) delivering 0.5% bupivacaine at 2mL/hr for 14 days was inserted above the incision and placed alongside the nerve into the Alcock canal (Fig. 5). The severed ends of the sacrotuberous ligament were reapproximated using a cadaver gracillis or Achilles tendon. Surgery was concluded by closure of the subcutaneous fat and overlying skin.

Primary outcomes were measured by assessing pain, comfortable sitting time, and global impression of change. Data were collected at the preoperative visit and at postoperative follow-up at 12 or 18 months. Pain was evaluated using an 11-point numerical rating scale of pain intensity. Patients were asked to rate their pain over a mean of 7 days. An individual change in pain score of ≥ 2 was regarded as clinically significant [5]. Comfortable sitting time was reported as the maximum amount of time the patient could sit before the urge to stand secondary to worsening pain. Global impression of change after surgery was measured by using a 6-point categorical scale. Patients were asked to describe their status after surgery as "much worse," "somewhat worse," "about the same," "somewhat better," "much better," or "cured."

The mean preoperative pain level was compared to the mean postoperative pain level using a 2-tailed *t* test for paired samples. The median preoperative sitting time was compared to the median postoperative sitting time using the Wilcoxon matched pairs signed rank test. For sitting times, we compared medians rather than means, because the sitting times were not normally distributed. For computational purposes, 1000 minutes was used for patients who reported indefinite sitting time. Global impression of change was correlated with comfortable sitting time by assigning each response a numerical value, sequentially from 3 to −2, starting with "cured."

Results

From June 2008 to March 2010, 10 consecutive patients (7 women and 3 men; age range, 29–81 years) underwent repeat operation with transgluteal decompression of the pudendal nerve. Location of neuropathic pain was unilateral (n = 8) or bilateral (n = 2). Location included a combination of clitoris or penis (30%), vulva or scrotum (70%), perineum (40%), and rectum (50%). Two patients also had additional pain outside the distribution of the pudendal nerve, i.e., the left buttock and abdomen, respectively. An inciting event was implicated in 9 patients, including pelvic surgery in 5, a fall in 2, and strenuous exercise in 2.

Outcomes from their initial surgery were categorized on the basis of history as "worsened" (n = 1), "no improvement" (n = 2), "some improvement but desired outcome not achieved"(n = 3), "initial positive outcome but now worsened"(n = 2), and "cured"(n = 2). Both patients that reported cure after surgery sustained subsequent injury that caused recurrence of pudendal neuralgia. Duration of symptoms before the first surgery ranged from 1 to 14 years (median, 2 years). Time to repeat operation was 1 to 7 years (median, 4 years).

Intraoperatively, adhesions were observed along the entire course of the pudendal nerve in 9 patients, and only at the ischial spine in 1 patient. In all patients, decompression of the pudendal nerve was technically successful (see "Materials and Methods"). There were no intraoperative complications. Postoperative complications included 1 wound infection that required incision and drainage.

One of the 10 patients was lost to follow-up after the initial postoperative visit. Of the remaining 9 patients, minimum follow-up time was 12 months (mean, 23 months; median, 25 months). Seven patients had at least 18 months of follow-up (Table 2).

Clinically significant improvement in pain was observed in 5 of 9 patients, with a change in the 11-point numerical pain scale of ≥ 2. Two patients reported complete resolution of pain, with a postoperative pain score of 0 at more than 18 months of follow-up. On average, the pain score decreased from 7.2 to 4.0 (p = .02).

Mean comfortable sitting time improved in 8 of 9 patients. Both patients with resolution of pain also reported complete resolution of pain on sitting. Overall, comfortable sitting time increased from a median of 5 minutes to 45 minutes (p = .008). One patient experienced no improvement, and continued to not tolerate any sitting.

Based on patient global impression of change, 89% of patients reported improvement of symptoms. Impression of change was described as "cured" (n = 2), "much

Table 2

Patient clinical findings and outcome

Age (yr)	Sex	Duration of Symptoms (yr)	Time to Repeat Operation (yr)	Outcome of Previous Surgery	Symptoms		Follow-up (mo)	NRS		Comfortable Sitting Time (min)		Global Impression of Change
					Side	Location		Preoperative	Postoperative	Preoperative	Postoperative	
81	F	10	5	No improvement	Bilateral	Vulva, Perineum, Rectum	14	10	10	0	0	About the same
64	F	1	1	Some improvement	Right	Vulva	14	10	3	10	180	Much better
64	F	3	2	Some improvement	Right	Clitoris, rectum	18	7	4	5	45	Much better
52	F	14	4	Worse	Left	Vulva	24	10	4	10	30	Somewhat better
44	M	1	4	Initially positive, but now worse	Right	Penis, perineum, scrotum, rectum	25	5	4	20	60	Much better
53	F	1	4	Cured, but with repeat injury	Left	Vulva, rectum	27	9	0	0	No limit	Cured
60	M	2	5	Initially positive, but now worse	Bilateral	Scrotum, perineum, rectum	28	6	6	5	30	Somewhat better
71	F	2	7	No improvement	Right	Vulva	29	5	5	5	20	Much better
47	M	1	2	Cured, but with repeat injury	Left	Penis, rectum	31	3	0	2	No limit	Cured

NRS = numerical rating scale of pain.

better"(n = 4), "somewhat better" (n = 2), and "about the same" (n = 1). No patient reported worsening of symptoms. Global impression correlated well with comfortable sitting time, with a correlation coefficient of 0.86 ($p < .05$).

Discussion

Pudendal neuralgia is a painful and debilitating neuropathic pain syndrome. Patients undergo a lengthy diagnostic process and treatment, which often results in a decision for surgical nerve decompression. However, in approximately 30% of patients, pudendal neurolysis may not provide relief of pain.

It is difficult to establish the reason for recurrent or persistent postsurgical pudendal neuralgia. Possible reasons for failure of surgery include new injury, incomplete decompression of the nerve, formation of postoperative adhesions with entrapment, and permanent nerve injury unresponsive to decompression. Outcomes of repeat operation may be explained by addressing these reasons for failure. Therefore, we believe that our modifications to the surgery contributed to successful outcomes.

Repeat surgery is much more technically difficult than the primary procedure. We chose a transgluteal approach for nerve decompression to offer the best exposure. The sacrotuberous ligament, which is the primary landmark used to localize the nerve, is transected or removed. The nerve is often encased in dense scar tissue, and there are no fat planes surrounding it. Use of a surgical microscope and nerve protective device enables us to find and directly visualize the nerve from the inferior edge of the piriformis muscle to the distal Alcock canal, permitting complete surgical decompression.

Continuous infusion of bupivacaine along the path of the nerve offers pain relief, with possible prevention of postoperative fibrosis. Bupivacaine inhibits fibroblast and myoblast growth [6]. In addition, application of the NeuraWrap Nerve Protector reduces adhesion formation around peripheral nerves operated on [7]. In patients who described initial improvement, return of symptoms may have been due to postoperative adhesions. Indeed, 9 of the 10 patients were found to have extensive adhesions encasing the nerve. Inasmuch as the NeuraWrap Nerve Protector and bupivacaine prevent adhesions, these patients may find long-term benefit after repeat surgery when the primary surgery has failed.

Platelet-rich plasma contains nerve growth factors that, in basic science studies, promote myelin growth [8,9] and improve wound healing in several applications [10]. Therefore, the platelet-rich plasma graft may be important in patients who do not experience improvement after decompression secondary to severe nerve damage.

Evidence demonstrates that the sacrotuberous ligament contributes to pelvic joint stability; nevertheless, the importance of its role or whether severing the ligament causes instability is debatable [11]. Although we have no objective measurements, repairing the sacrotuberous ligament may improve patient pelvic stability. Physical examinations by

our physical therapist have revealed increased joint mobility in patients with a severed sacrotuberous ligament.

In the present study, 89% of patients reported improvement in symptoms, with 2 patients reporting complete resolution of pain. Both of these patients reported cure after the primary surgery, but sustained another injury and recurrence of pudendal pain. This outcome can be explained by treating recurrence of compression.

Our outcomes are supported by an 11-point numerical pain scale and comfortable sitting time that was recorded independently without recall bias. Although only 55% of patients reported improvement of pain, the improvement of comfortable sitting time better correlated with patient perception of surgical outcome. Indeed, in a condition where sitting is limited by pain, improving the capacity to sit comfortably reduces the effect of the disease and leads to overall well-being.

The present study demonstrates that patients with persistent pudendal neuralgia after surgical decompression or those who sustain another injury may benefit from repeat operation with our modifications. Conclusions of this study are limited secondary to small sample size, and a prospective study should be performed. Nevertheless, for an uncommon condition with limited treatment options, repeat operation may be considered in patients with persistent pudendal nerve entrapment.

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