Diagnosis and treatment of pudendal nerve entrapment syndrome subtypes: imaging, injections, and minimal access surgery

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Object. To improve diagnostic accuracy and achieve high levels of treatment success in patients with pudendal nerve entrapment (PNE) syndromes, the author of this study applied advanced technology diagnostics in distinguishing the various syndrome types according to the different entrapment locations and evaluated new minimal access surgical techniques to treat each subtype.

Methods. Two hundred cases were prospectively evaluated using a standardized set of patient-completed functional and symptom assessments, a collection of new physical examination maneuvers, MR neurography, open MR image-guided injections, intraoperative neurophysiology, minimal access surgery, and formal outcome assessment with the Oswestry Disability Index, pain diagrams, and analog pain scales.

Results. Four primary types of PNE syndromes were identified based on the different locations of entrapment: Type I, entrapment at the exit of the greater sciatic notch in concert with piriformis muscle spasm; Type II, entrapment at the level of the ischial spine, sacrotuberous ligament, and lesser sciatic notch entrance; Type III, entrapment in association with obturator internus muscle spasm at the entrance of the Alcock canal; and Type IV, distal entrapment of terminal branches. The application of new, targeted minimal access surgical techniques led to sustained good to excellent outcomes (50–100% improvement in the pain score or functional score) in 87% of patients. Most of these patients obtained most of their improvement within 4 weeks of surgery, although some continued to experience progressive improvements up to 12 months after surgery.

Conclusions. The application of advanced diagnostics to categorize PNE syndrome origins into 4 major subtypes and the subsequent treatment of each subtype with a tailored strategy greatly improved therapeutic outcomes as compared with those reported when only a single treatment paradigm was applied to all patients. (DOI: 10.3171/FOC.2009.26.2.E9)

KEY WORDS • magnetic resonance neurography • nerve decompression • neuroplasty • open magnetic resonance imaging-guided injection • outcome study • pudendal nerve entrapment

Although entrapments affecting the distal peripheral nerves, such as carpal tunnel syndrome and cubital tunnel syndrome, are well understood by virtually every neurosurgeon and neurologist, there has been little interest in perineal neurology and neurosurgery. Urologists and gynecologists are focused on end organs so that neurological conditions affecting the perineum have been effectively relegated to orphan status. Entrapment of the pudendal nerve is an area with extremely few practicing specialists.

The medical and surgical management of PNE syndrome has an extended history, but no rigorous studies with significant patient numbers were published until 1998. And although there are now a number of reports covering various aspects of diagnosis and treatment, the success rate in treating pudendal neuralgias has been limited. One problem has been that there appear to be several subtypes with different anatomical sites of entrapment and clinical presentations. Furthermore, the surgical anatomy of the pudendal nerve itself is complex (Fig. 1) and subject to significant individual variation.

Targeting of the pudendal nerve for diagnostic or therapeutic injection at the ischial spine by using an electrodiagnostic technique, C-arm fluoroscopy, and CT scanning has been described. Diagnosis has also relied on pudendal nerve latency testing, but imaging has played very little role in diagnosis. Recently, advances in

Abbreviations used in this study: FFE = fast field echo; FSE = fast spin echo; PNE = pudendal nerve entrapment; SPIR = spectral presaturation inversion recovery; TSE = turbo spin echo.
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clinical nerve imaging in the form of MR neurography,15–17 as well as open MR image–guided injections,11 and the development of new minimal access surgical techniques for deep pelvic nerve entrapments13 have opened the possibility of improving the specificity and success of treatments for PNEs.

Methods

This study was based on the prospective application of an evaluation program in 200 consecutive patients who had presenting signs or neurological symptoms potentially referable to the pudendal nerve distribution—typically including pain, numbness or dysfunction in the perineum, low medial buttock, genitalia, rectum, or proximal medial thigh, or dysfunction of the rectal or urogenital system associated with pain or numbness and for which no organic urological, gynecological, or proctologic cause could be determined by prior specialist evaluation.

Evaluation in all patients included a detailed neurological physical examination and diagnostic MR neurography using protocols described previously.15 Open MR image–guided injections were performed for diagnosis with either a Siemens or Philips interventional MR system in patients in whom the physical examination and imaging studies had suggested a specific testable diagnosis.

Treatments included open MR image–guided injections of Marcaine, steroids, Botox, and hyaluronidase as well as minimal access surgical decompressions with the intraoperative placement of adhesiolytic agents, typically Seprafilm. Outcome monitoring involved the completion of Oswestry Disability Index (version 1) questionnaires, pain diagrams, and analog pain scales at the time of the initial visit, at the time of maximum response during the 2-week interval following any diagnostic intervention, and at 2, 4, and 12 weeks after surgical treatments. One-year posttreatment data were sought from patients as well.

Physical Examination

An expanded physical examination for pelvic entrapments was performed as follows in all patients. The patient was initially examined in a sitting position and tested for thigh elevation, depression, abduction, and adduction against resistance; this evaluation attended to strength as well as the potential to elicit or reproduce symptoms. Major proximal leg weakness drew attention to high lumbar roots or spinal nerves, but adductor pain or weakness reflected obturator nerve impingement either adjacent to the obturator internus muscle or at the obturator foramen.

The patient was then asked to lie supine, and each leg was assessed for the reproduction of symptoms through a straight leg raise, passive internal and external rotation of the leg with the hip and knee flexed, and resisted abduction and adduction of the flexed internally rotated thigh.13 To check for the relief or exacerbation of symp-
toms, each leg was subject to “crossed leg traction,” with the examiner standing at the foot of the bed and pulling the leg upward ~ 10°, toward the contralateral side ~ 10°, and applying traction by pulling at the ankle. While lying supine with his or her leg extended, the patient was asked to elevate the leg against resistance applied at the knee. The area along the inguinal ligament was palpated with specific attention to the obturator foramen.

Hip or groin pain with passive hip rotation signaled the possibility of a primary hip joint pathology, whereas the reproduction of symptoms by resisted adduction or abduction as well as relief by crossed leg traction indicated the possible involvement of the piriformis or obturator internus muscles. Note that patients who have been engaged in a directed program of piriformis muscle stretches often test falsely negative for these maneuvers. The aggravation of symptoms on elevating the extended leg but not while elevating the flexed leg (lying supine vs sitting) was indicative of possible pathology involving the psoas muscle. Inguinal palpation was meant to identify direct sensitivity referable to the ilioinguinal or genitofemoral nerves. Isolated obturator foramen tenderness was believed to indicate obturator internus muscle spasm.

The patient was asked to step down from the examination table and stand. Working from behind the patient, the lumbar, sacral, and coccygeal spine was palpated. This part of the evaluation was performed to differentiate regional spinal pains and check for the possibility of a pelvic floor muscle spasm that can present as coccygeal pain.

In the pelvis, the upper buttock just below the iliac crest was palpated for tenderness to distinguish low-back pain from gluteus maximus pain (superior gluteal nerve distribution). Palpation over the posterior superior iliac spine as well as medial and lateral to it can reveal an L-5/S-1 facet syndrome, sacroiliac joint pain, or superior cluneal nerve involvement, respectively.

The greater trochanter (for bursitis) and the area superior and inferior to the trochanter (tensor of the fascia lata, which is also superior gluteal nerve innervated) were examined. The sciatic notch was palpated for tenderness (piriformis muscle pain) with attention also directed to the sacroiliac ligament superomedially and the sacrotuberous ligament inferomedially. The sacroiliac ligaments are subject to direct tears, and in some patients the pudendal nerve is adherent to the deep surface of the sacrotuberous ligament. It is helpful to know if direct palpation of these ligaments aggravates pudendal referable symptoms. An area inferior and just medial to the greater sciatic notch was palpated to assess the inferior retrosciatic area for tenderness related to the ischial spine and sacrospinous ligaments, which are possible sites of PNE.

Palpation was performed on the lateral aspect of the ischial tuberosity for ischial tunnel syndrome—which can involve local adhesion, adhesions of the posterior femoral cutaneous nerve or sciatic nerve to the hamstring origin, or aggravation of these nerves by the transiting obturator internus tendon when it is under abnormal tension—directly on the ischial tuberosity for bursitis or inferior cluneal nerve involvement, and on the medial aspect of the ischial tuberosity by reaching from between the legs to assess obturator internus muscle tenderness (Fig. 2).

The patient was asked to bend forward at the waist to screen for lumbar disk pathology, to extend the back to screen for a facet syndrome, to twist the spine to the left and right to assess for lumbar annular tears, and to perform lateral bends to the right and left, which can aggravate symptoms from the gluteal or piriformis muscles when bending toward the side ipsilateral to the pathology. Finally, a Romberg test was conducted, and the patient was asked to walk heel to toe along a line to check for muscle- or nerve-based hip instability as a cause of impaired walking balance in the setting of normal cerebellar and spinal cord function.

**Magnetic Resonance Imaging Examination**

For patients in whom palpation of the pelvis produced unclear findings or seemed to cause pain in atypical locations (for example, the ischial margin or points lower, higher, more medial, or more lateral than typically encountered), an open MR image study was performed. For this procedure, a 0.23-T open-configuration interventional Panorama system (Philips Medical Systems) was used, and the patient was placed prone or in a lateral decubitus position as appropriate. Using a magic marker, a mark was made on the midbuttock at the approximate level of the tip of the greater trochanter, and the laser-centering system was used to define this mark as the midpoint of the image. A T1-weighted multislice axial acquisition was performed (4 minutes 30 seconds), and data were displayed in the lower right of 4 image panels on the in-room monitor.

The examiner, sitting next to the patient, identified the major points of anatomy by obtaining a series of T1-weighted FFE MR images (~ 12 seconds each) while deeply palpating various structures with his finger. The
3 images—a center image, another 1 cm superior to the center, and the other 1 cm inferior to the center—were displayed on the remaining 3 panels of the 4-panel in-room monitor display. Additional skin markings were made in this way to identify the location of the sciatic notch, ischial tuberosity, and greater trochanter.

The patient was given an opportunity to point to areas of pain while the 12-second T1-weighted FFE MR images were obtained and while the examiner performed a series of palpation and imaging steps (Fig. 3). When the precise location of greatest sensitivity remained unclear, the examiner pressed on 3 adjacent locations and asked the patient to specify which of the 3 was the most sensitive. Thus, points of sensitivity could be defined in terms of specific anatomical structures in the deep posterior pelvis.

**Magnetic Resonance Neurography**

Magnetic resonance neurography images of the pelvis were obtained as previously described.15-18 The FSE or TSE images were obtained in 1.5-T units (GE Medical Systems, Siemens, or Philips Healthcare) by using chemical shift selection, inversion recovery, or SPIR for fat suppression. Gradients were ≥ 10 mT/m. In each case, the magnet was reshimmed while the patient was in position before commencing data acquisition. Commercially available phased array coils were used to enhance signal-to-noise performance. For pudendal nerve imaging, echo train length parameters were reversed relative to those for sciatic imaging; an echo train length of 4 was applied to optimize spatial resolution in the coronal plane in which the cross-sections had been obtained, but an echo train length of 8 was used for longitudinal pudendal nerve images in the axial planes. The field of view was minimized for each study. In all diagnostic MR neurography studies, T1-weighted spin echo and FSE or TSE images were collected. For the FSE images, the TE was 95–110 msec, TR (time to repeat) 4–5000 msec, number of excitations 2–4, and resolution 256 × 256–512 × 512. In FSE inversion recovery imaging the TE was 50 msec, and in SPIR imaging it was 75 msec. Slice thickness was 3 mm with 0-mm spacing. The acquisition of axial T1-weighted MR images was followed by T2-weighted fat-suppressed imaging in the axial and coronal planes. For patients with a strong component of sciatic involvement, additional T1- and T2-weighted fat-suppressed images were acquired in the plane perpendicular to the course of the sciatic nerve where it traverses the sciatic notch. All neurographic images were obtained with a TE > 40 msec to eliminate any potential “magic angle effects.” Images were then subjected to multiplanar reformat postprocessing in eFilm (Merge Healthcare) with particular attention devoted to the course of the pudendal nerve as it descends from the ischial spine to enter the canal of Alcock on the medial aspect of the obturator internus muscle.

**Open MR Image–Guided Injections**

Open MR image–guided injections were performed in a Siemens 0.25-T Concerto or Viva interventional MR imaging system or a Philips 0.23-T Panorama interventional system with Optiguide—both outfitted with in-room monitors. Twenty two–gauge, 15-cm titanium Lufkin needles (EZ-EM) were used for the injections. Imaging commenced with a multislice anatomical T1-weighted image of the pelvis for positional reference and included a series of T1-weighted FFE 12-second (Philips) or fast low-angle shot (FLASH) 16-second images (Siemens). Additional fast T2-weighted 5- or 7-slice series were obtained as needed during the course of the procedure to provide optimized detail of the spread of the injected agent. Patients were prone in the Siemens Concerto or Viva systems and in a lateral decubitus position in the Philips Panorama system; the latter position was possible because of the larger gap between magnet poles in that system. Injections in the piriformis muscle consisted of 12 ml of 0.5% bupivacaine and 1 ml of Celestone Soluspan (betamethasone acetate and betamethasone phosphate injectable suspension, 6 mg/ml). Injections in the obturator internus muscle were 6 ml of 0.5% bupivacaine and 0.5 ml of Celestone Soluspan. When a pudendal block was indicated at the ischial spine or along the medial aspect of the obturator internus muscle at the entrance to the Alcock canal, 3 ml of 0.5% bupivacaine and 0.5 ml of Celestone Soluspan were used. When Botox (botulinum toxin, Type A) was administered in a muscle, the volume of Marcaine was reduced by 50% and was administered as 0.75% bupivacaine—preservative free to minimize the risk of denaturing the injected protein. When hyaluronidase (Amphadase, 150 U/ml) was administered along a nerve, the bupivacaine was similarly adjusted. For Botox, 100 U was administered in 6 ml of preservative-free saline in the piriformis muscle and 3 ml in the obturator internus muscle. A second dose of 100 U was administered when a bilateral injection was required. Hyaluronidase was administered as 300 U in 2 ml of preservative-free saline. All Botox and hyaluronidase injections were administered simultaneously with the bupivacaine and Celestone Soluspan at the same site.

Botox and hyaluronidase proved most useful for man-
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aging minor postoperative recurrences. However, these agents were also used for second injections in 4 patients who had prolonged relief (> 2 weeks) following bupivacaine andCelestone Soluspan injection into the obturator internus and piriformis muscles.

In selected patients with no alteration in symptoms from dense pudendal nerve block and who had good confirmation of no other cause for the symptoms, open MR imaging was used to guide injections via a lateral subcoccygeal approach to block the ganglion impar with 3 ml of 0.5% bupivacaine and 0.5 ml of Celestone Soluspan to rule out autonomically driven, nonpudendal saddle area pain syndromes. This approach was similar to previously reported paramedian approaches39 as opposed to the transcoccygeal approaches.31,41 Blocks of the inferior cluneal branches of the posterior femoral cutaneous nerve, of the ilioinguinal/genitofemoral nerves, and of the obturator nerve were also used to rule out alternative somatic sources of pain symptoms in some patients.

Because of the large volume of bupivacaine necessary, all procedures were conducted in an open MR imaging surgicenter setting with available MR imaging–compatible anesthesia and resuscitation equipment. Aspiration was performed after each injection of 2 ml of bupivacaine to minimize the risk of respiratory or cardiac compromise due to intravascular injection. The needle was repositioned if the injected agent did not spread evenly in the muscle or if any leakage from the muscle was observed. Fast T2-weighted MR images in the coronal, sagittal, and axial planes were acquired to verify good distribution of the injected agent. During each 30-minute imaging procedure 15–25 image series were typically obtained. Full multislice postinjection T2-weighted MR images were used to assess the final distribution of the agent injected in the muscle, and these images were also read for diagnostic information on the degree of muscle spasm or fibrosis at the injection site.

Surgical Technique

Surgeries were generally conducted using minimal access approaches on an outpatient or overnight-stay basis. Those involving piriformis muscle resection had the longest recovery times, which lasted up to 2 weeks. Most patients had only a few days of mild postoperative pain. All patients were mobilized immediately after surgery but were restricted from heavy lifting for a 3-month postoperative period.

Intraoperative electromyography monitoring of muscles innervated by the pudendal nerve, the tibial and peroneal components of the sciatic nerve, the nerve to the obturator internus muscle, and the inferior and superior gluteal nerves was used for nerve identification and protection in all patients. Some surgeries were performed using real-time intraoperative open MR image guidance, but most were conducted in a standard operating room with the aid of immediate preoperative radiographic localization and intraoperative nerve stimulation.

Depending on the presentation and subtype of syndrome in an individual patient, 1 of 4 different surgical procedures were required: 1) a superior transgluteal approach for piriformis muscle resection, and 1a) a superior retrosciatic dissection for neuroplasty of the pudendal nerve and obturator internus nerve in the greater sciatic notch, and/or 1b) an inferior retrosciatic dissection for neuroplasty of the nerve to the obturator internus muscle and the pudendal nerve in the area of the ischial spine and entrance to the lesser sciatic notch and the Alcock canal entrance; 2) a medial translgluteal approach for access to the pudendal nerve and the nerve to the obturator internus muscle at the greater sciatic notch, the ischial spine, the lesser sciatic notch, and the entrance and full length of the Alcock canal; 3) an inferior translgluteal approach for access to the sacrotuberous ligament, the full length of the Alcock canal, and the retrosciatic space; or 4) a transischial approach for access to distal branches of the pudendal nerve.

Superior Transgluteal Approach. The patient is positioned prone on bolsters so that the knee falls below the level of the hip, which provides relative elevation of the greater trochanter at the surgical site and thus aiding access to the piriformis tendon. Placement of a 3-cm incision is based on locating the superior medial edge of the greater trochanter of the femur on a posteroanterior hip radiograph. Note that the length and orientation of the femoral neck as well as the size of the greater trochanter can vary significantly among individuals.

Progress is made through the subcutaneous adipose tissue using a Bovie monopolar coagulator (Valleylab, Tyco Healthcare), but this device is not used at any point after reaching the gluteal fascia. The gluteal fascia is opened with Malis bipolar cautery (Codman/Johnson & Johnson) and Metzenbaum scissors; blunt finger dissection through the leaves of gluteal musculature minimizes exposure trauma and helps to ensure outpatient management. Exposure is maintained with a Shadowline retractor system (V. Mueller), an anterior cervical type retractor with a blade/retractor connection providing good rigidity deeply under strong tension, as well as a release lever system that allows for rapid, controlled, atraumatic replacement of blades as the depth of surgery progresses. A set of blades up to 100 mm in length (custom made by V. Mueller) is sufficient for nearly all patients, although longer Omni blades may be required in extremely heavy patients.

Safety for the sciatic nerve is accomplished by carefully progressing through the muscle layers until the hard, clear piriformis fascia is reached with dense yellow fat behind it. Older techniques in which a Bovie coagulator is used to slice through gluteal muscles pose a grave risk to the major nerve elements and should never be used; they provide no view ahead of the destructive cutting energy and lead to prolonged or permanent gluteal muscle dysfunction.

Once the piriformis fascia is brought into view by gentle blunt dissection, the retractor blades are reset, and the fascia is opened carefully with bipolar cautery and Metzenbaum scissors. An electrodiagnostic system (set at 0.5–10 mA) with electromyography monitoring of multiple muscles innervated by the superior gluteal, inferior gluteal nerve, tibial nerve, and peroneal nerve is used to identify nerves prior to their exposure in the piriformis fat pad. When the nerve being sought is not immediately
visible, a high milliamperage is used to locate its vicinity, and decreasing milliamperage is then applied as the dissection approaches the nerve. In this fashion, it is possible to locate and protect the sciatic, inferior gluteal, and superior gluteal nerves with a high degree of reliability and safety.

The sciatic nerve is partially mobilized. Using a view of the location of the superior surface of the sciatic nerve, together with palpation of the greater trochanter and the sciatic notch, the borders of the piriformis muscle are identified and confirmed. There is a considerable range of variation in the anatomy of the piriformis muscle and its relation to the sciatic nerve so that full identification, orientation, and protection are essential before any resection is commenced. The orientation of the femoral neck relative to the femoral shaft varies from nearly horizontal to nearly vertical, and the height of the greater trochanter is variable as well. In a patient with a horizontal femoral neck and large greater trochanter, the piriformis muscle can be nearly perpendicular to the sciatic nerve. In contrast, in a patient with a vertical femoral neck and short greater trochanter, the piriformis muscle is nearly parallel with the sciatic nerve.

Many patients have multipartite piriformis muscles, and in some the superior border appears fused with deep gluteal muscles. Some patients have an accessory piriformis muscle compressing the more proximal portion of the sciatic nerve, and this accessory muscle is sectioned and removed as well. Special attention and caution is required in patients in whom preoperative imaging has demonstrated a split piriformis muscle traversed by a split sciatic nerve. The slip of muscle passing between the tendo calcaneus and peroneal sciatic components must be removed as well. It is always helpful to ensure with electrodiagnostic stimulation that both the tendo calcaneus and peroneal sciatic component muscles are identified and confirmed. There is a considerable range of variation in the sciatic nerve so that it is soft and pliable as it is pushed together with palpation of the greater trochanter and the sciatic notch, the borders of the piriformis muscle are identified and confirmed. There is a considerable range of variation in the anatomy of the piriformis muscle and its relation to the sciatic nerve so that full identification, orientation, and protection are essential before any resection is commenced. The orientation of the femoral neck relative to the femoral shaft varies from nearly horizontal to nearly vertical, and the height of the greater trochanter is variable as well. In a patient with a horizontal femoral neck and large greater trochanter, the piriformis muscle can be nearly perpendicular to the sciatic nerve. In contrast, in a patient with a vertical femoral neck and short greater trochanter, the piriformis muscle is nearly parallel with the sciatic nerve.

One or 2-O silk ties are placed around the muscle for ongoing identification and control. Bipolar cautery and Metzenbaum scissors can be used to fully transect the muscle in 2 locations with ongoing complete hemostasis. Injection of an anesthetic agent into the muscle immediately prior to resection can help minimize postoperative discomfort. Removing an ~2-cm-long segment of muscle helps to prevent readhesion of the separated segments, which can occur when only a single cut is made. This procedure also generally entails severing the small nerve to the piriformis muscle, resulting in subsequent atrophy of any remaining components.

Neuroplasty of the distal lumbosacral plexus, sciatic nerve, and posterior femoral cutaneous nerve is then performed by blunt dissection generally by using DeBakey pickups and a tonsil clamp. Specifically, any abnormal fibrous covering is separated from the nerve so that the nerve is free and fully mobile at the end of the dissection. In many cases, fibrovascular bands cross or compress the sciatic nerve and can be cut. A gentle dissection technique, the liberal use of electrodiagnostic stimulation when nerve locations are in question, and meticulous hemostasis with bipolar cautery before cutting any tissue promote the safety of neural tissues.

By swinging the retractor system, ready access to the sciatic nerve can be achieved from the top of the ischial tuberosity to well inside the sciatic notch allowing full mobilization of at least 12 cm of the nerve course. Extensive muscle resection inside the pelvis along its sacral aspect inside the sciatic notch is not recommended on a routine basis because of a higher risk to autonomic fibers in the presacral area.

Once the piriformis muscles is resected and any fibrous bands restricting the sciatic nerve inside the sciatic notch are released, the sciatic nerve can be mobilized and gently lifted superiorly for a superior retrosciatic dissection to access the obturator internus and pudendal nerves, which are deep to the sciatic nerve at this location and run between the sciatic nerve and the osseous ischial margin of the sciatic notch. The pudendal and obturator internus nerves can be identified using electrodiagnostic stimulation and brought into view as necessary. Any fibrovascular bands or adhesions affecting the pudendal nerve and the nerve to the obturator internus muscle in this area can now be released. Large veins or arteries adherent to nerves can be carefully separated from them, but extensive ligation or coagulation of significant vessels should be avoided so that the expansion of newly recruited vessels, which can cause symptoms, does not occur postoperatively.

Seprafilm should be cut into small squares of ≤1 cm and dipped into a small pool of irrigation fluid inside the surgical site immediately before inserting it behind the sciatic nerve so that it is soft and pliable as it is pushed into place in multiple layers. Seprafilm will not adhere to a cottonoid, although it will stick to metal instruments. Two standard sheets of Seprafilm are usually required to cover all surfaces.

Dexamethasone (10 mg) can be administered intravenously at the start of the procedure. Powder-free gloves must be used by both the surgeon and any surgical tech who touches the instruments or the Seprafilm to further reduce the risk of postoperative fibrosis. Only bipolar cautery is used once the gluteal fascia is reached. Meticulous and complete hemostasis must be confirmed prior to closure. On completion of the neuroplasty, the wound should be irrigated copiously with antibiotic irrigant fluid maintained at body temperature in a solution warmer. Seprafilm pieces should be placed in layers on all dissected nerve surfaces as an adhesiolytic agent after extensive irrigation to avoid washing out the hyaluronate, which is the active agent carried by the Seprafilm.

Bupivacaine (0.5% without epinephrine) is applied to the Seprafilm and dissected nerves and is instilled in gluteal muscles along the line of approach. Epinephrine must not be used in an anesthetic agent applied directly to nerves in this setting to avoid the possibility of vasocostriction causing nerve ischemia. The gluteal fascia is closed with 1-0 Vicryl sutures. No drain is placed. The skin is sutured with a closely placed series of interrupted 3-0 Vicryl sutures because of the mechanical stress on this body region. A subcuticular stitch of 4-0 Vicryl and the application of cyanoacrylate as well as Steri-Strips over skin adhesive promote wound healing. Patients are allowed to ambulate immediately. Those who experience
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significant muscle spasm or local pain are provided with pain management in the facility overnight.

Retrosciatic Dissection. Following the approach to and any resection of the piriformis muscle, a retrosciatic dissection is commenced. Using an approach over the posterior and inferior surface of the sciatic nerve and relying on intraoperative stimulation, the nerve to the obturator internus muscle is identified and tracked proximally into the greater sciatic notch where it runs deep to the sciatic nerve. Neuroplasty of the pudendal nerve from this point and continuing inferiorly across the ischial spine and into the lesser sciatic notch is then performed. From this access, it is generally possible to dilate the Alcock canal to a depth of 2–3 cm along its course by gentle application of a long tonsil clamp. Of course, one carefully avoids disrupting the veins and arteries in the inferior retrosciatic space, but any bleeding should be managed initially with gentle pressure and the placement of fibrin-soaked Gelfoam to avoid excessive coagulation near the numerous nerve elements.

Medial Transgluteal Approach. A medial transgluteal approach with a 3-cm incision centered over (directly posterior to) the midpoint of the acetabulum is used in patients with ischial spine or Alcock canal syndromes without piriformis muscle involvement on physical examination. This approach requires careful identification of the sciatic nerve and then a direct retrosciatic dissection to identify and track the obturator internus and pudendal nerves. The more medial and inferior position of this incision allows greater access to the sacrotuberous and sacrospinous ligaments. Most patients with PNE at the level of the ischial spine have a very medially placed pudendal nerve that often adheres to the anterior surface of the sacrotuberous ligament. The nerve is identified electrodiagnostically and carefully mobilized from the sacrotuberous ligament so that the ligament can be partially resected. Some patients with entrapment in this location have a variant fibrous septum sealing the entrance to the lesser sciatic notch that is perforated by the nerve. This septum should be carefully opened or resected to improve the mobility of the nerve as it enters the lesser sciatic notch. In some patients, the sacrospinous ligament may need to be partially sectioned if the edge impinges on the nerve at this location.

This medial transgluteal approach also offers excellent, magic angle access along the Alcock canal because the line of approach is parallel to the canal. Thus, it is possible to noninvasively access the canal to dilate it, relying on careful electrodiagnostic monitoring to accurately track its course along the medial aspect of the obturator internus muscle.

Inferior Transgluteal Approach to the Sacrotuberous Ligament. A 3-cm incision is centered between the ischial spine and ischial tuberosity as revealed on an initial posteroanterior pelvic radiograph with markers. This approach takes the surgeon directly to the sacrotuberous ligament. Once the ligament is exposed, the pudendal nerve can be accessed distally along the course of the Alcock canal as well as its proximal approach to the lesser sciatic notch. In patients in whom the pudendal nerve densely adheres to the deep surface of the sacrotuberous ligament, it is possible to partially or completely resect the ligament to access the nerve as some have advocated, but most entrapments can be released without sectioning this major structural ligament.

Transischial Approach. In patients with distal branch entrapments (for example, isolated penile numbness), a transischial approach can be used for minimal access decompression. The incision is made parallel and just medial to the ischial tuberosity. In this fashion, the pudendal nerve can be identified electrodiagnostically on the medial aspect of the obturator internus muscle, and there is good access to track and decompress the rectal branch of the pudendal nerve as it proceeds medially or the distal genital branches as they proceed anteriorly. Hruby and colleagues have also described direct anterior approaches for the decompression of small distal sensory branches of the pudendal nerve.

Results

Presenting Factors

The symptoms of PNE involved pain, dysfunction, and numbness in all or part of the distribution of the nerve, that is, the saddle area between the legs including the genitalia, rectum, and terminal urinary tract. In addition, sexual and sphincter dysfunctions were seen. Sexual dysfunctions included female continuous arousal as well as an absence of sensation, male impotence, and dyspareunia. Aggravation by sitting was common when the obturator internus or piriformis muscles were involved, but there was not always a positional trigger. The onset of symptoms could be insidious with no clear precipitant, but a history of local trauma or a pulled muscle in the pelvis was also common. Bicycle riding was also a common historical factor in patients with involvement of the obturator internus muscle.

Unlike other peripheral nerve disorders, pudendal syndromes are often bilateral. Indeed, more than one-half of the patients in this study reported bilateral symptoms. Nonetheless, a purely symmetrical set of neurological symptoms with no focal sensitivity to palpation at the piriformis muscle, obturator internus muscle, or area of the ischial spine and for which there are no positional aggravating factors presents a very low likelihood of an anatomical diagnosis or surgical treatment success.

Patients ranged in age from 8–82 years. There was no significant male or female preponderance. Apparently, because many physicians lack familiarity with this syndrome, the time to diagnosis after the onset of symptoms was often > 2 years and occasionally even > 10 years. Most patients had seen more than 5 physicians from specialties including urology, gynecology, general surgery, psychiatry, neurosurgery, neurology, and pain management. Failed prior interventions included artificial lumbar disk placement, lumbar fusion, sacroiliac joint fusion, hysterectomy, prostatectomy, rectal surgery, and a wide variety of nonspecific pain treatments such as acupuncture and trigger point injections.
Physical Examination Findings

On physical examination, > 95% of patients had findings directly related to a potential PNE syndrome. The most common finding was sensitivity to palpation at the obturator internus muscle that was elicited by manual compression on the deep medial aspect of the ischial tuberosity. Other findings included sensitivity to palpation in the sciatic notch, the greater trochanter of the femur, and the ischial margin at the inferior aspect of the sciatic notch. Reproduction of symptoms with straight leg raises was rare; however, adduction of the thigh in a seated position, passive internal or external rotation of the hip joint, and resisted abduction or adduction of the flexed internally rotated thigh elicited symptoms in many individuals.

Diagnostic Test Results

Findings on electrodiagnostic testing of pudendal nerve latency with or without provocative positioning was abnormal in some patients, although the test was performed and findings were positive in only a small subset of patients in this study. These data were reviewed in patients who had undergone this test elsewhere, but the test was not administered as part of the present study. There was no correlation between negative or positive results on this test, and an accurate diagnosis and successful treatment. Note, however, that the present study did not constitute a formal evaluation of that test.

Magnetic resonance neurography provided detailed views of relevant anatomy throughout the course of the pudendal nerve (Fig. 4), and findings included asymmetry of the piriformis or obturator internus muscles. Moreover, asymmetric swelling or deformation of the pudendal neurovascular bundle in the Alcock canal was often seen (Fig. 5). Edema of the pudendal neurovascular bundle at the ischial spine was also observed on MR neurography studies in patients with that subtype of the syndrome (Fig. 6).

Unlike patients in an MR neurography study in the setting of sciatica of nondisc origin, the population in the present study was highly selected by referral. In the sciatica study, MR neurography was performed on a blinded basis in a group of patients identified from the UCLA Comprehensive Spine Center whose routine spi-
neuronal diagnostics and treatments had failed and whose outcomes were matched with those in a previous long-term study of identically selected patients. Thus, patients with an affected piriformis muscle constituted only a fraction of the total number enrolled, and their positive findings on MR neurography could be reliably identified with regard to false-positive and false-negative rates so that accurate and meaningful sensitivity and specificity could be identified. Assessing the sensitivity and specificity of MR neurography in the sciatica–piriformis study was also more readily feasible because there are other well-known and readily diagnosed causes of sciatica.

A formal comparison of the MR neurography findings in the present study's patient population with pudendal symptoms and those of a matched group with piriformis syndrome only will be reported in a separate paper. However, the current study reveals a number of highly specific findings that are well revealed on MR neurography. Images in patients with distal pudendal entrapments not associated with obturator internus muscle spasm typically showed dilation and increased image intensity affecting the pudendal nerve in its course along the Alcock canal. The images in a number of these patients also demonstrated anatomical abnormalities or atrophy of the obturator internus muscle on the involved side.

All patients in whom obturator internus spasm had been diagnosed in this study also had vessel dilation on the medial aspect of the obturator internus muscle posterior to the entrance of the Alcock canal. This finding was also typically correlated with the side involved (ipsilateral or bilateral); that is, a patient with unilateral obturator internus muscle spasm typically had unilateral vessel dilation.

Because MR neurography is generally a very sensitive test, negative studies—those with no dilation of the pudendal nerve or associated vessels on the medial aspect of the obturator internus muscle and no hyperintensity of the pudendal nerve at the ischial spine or along any other part of its observable course—were very useful in diagnosis. These negative imaging studies are comparable to a completely normal-appearing lumbar MR image obtained in a patient with back or leg pain. Patients with symptoms possibly referable to the pudendal nerve but in whom there was no confirmatory finding on physical examination were scheduled to undergo MR neurography. If the study was positive—typically showing a very distal pudendal nerve hyperintensity—then an injection procedure was planned.

If both the MR neurography study and the physical examination were negative, the diagnostic workup was considered negative for PNE. In this situation, after re-evaluation for possible inflammatory, autoimmune, or end-organ pathology, an MR image-guided injection of the ganglion impar was then performed. If the ganglion impar injection also failed to affect the pain or if a series of 2 ganglion impar blocks failed to provide persistent relief of the syndrome, then referral to a pain specialist for consideration of an epidural or sacral root stimulator was suggested. With this subset of patients, it is particularly important to be confident that all potential end-organ causes of the symptom have been properly evaluated. Further imaging with standard MR neurography is unlikely to provide additional information at that point. In some situations, diffusion weighted or diffusion tensor imaging can be performed with the aid of relevant tractography analysis software to distinguish small nerve branches from similarly shaped surrounding structures, which may prove helpful in resolving these difficult cases in the future.

In the highly selected group of patients referred to this practice, negative MR neurography and physical examination findings were infrequent and occurred in only 5.5% of the patients in the present study. Ganglion impar syndromes successfully resolved with injections accounted for 3 of 11 patients with otherwise nondiagnostic workups.

Open MR image-guided injections offered excellent access to the pudendal nerve at a variety of locations not readily seen on radiography-based guidance modalities such as fluoroscopy and CT (Fig. 7). Magnetic resonance imaging also avoided unnecessary pelvic irradiation with x-rays and provided far greater safety (for example, avoiding puncture of adjacent bowel structures) as compared with electrodagnostic guidance.

Although many patients report bilateral symptoms, such sensations often reflect the bilaterality of the neurological sensory representation of perineal midline structures. In these patients, unilateral blocking on the side associated with physical examination findings often produces bilateral relief.

Syndrome Subcategories

Overall diagnostic efficacy based on patient history and specific positive and negative results on physical ex-
amination, imaging, and injections was > 95%, leaving only a small number with symptoms neurologically consistent with PNE but no detectable findings or responses to diagnostic tests in the highly selected group referred to this practice. Most important was the discovery that PNE can be categorized into 4 major categories based on the location of the entrapment. Selectively directing the various minimal access surgical treatments at these different syndrome subtypes greatly improved the overall success rate for achieving immediate and lasting relief of presenting symptoms, which occurred at a rate generally comparable to those previously reported for the management of pelvic sciatic nerve syndromes.13

Pudendal nerve entrapment categories were identified as follows among the 189 patients in whom PNE was diagnosed (excluding 11 patients who presented with symptoms suggestive of PNE but whose evaluation was nondiagnostic, as outlined in the text): Type I, entrapment exclusively at the level of the piriformis muscle in the sciatic notch only (4 patients [2.1%]); Type II, entrapment at the level of the ischial spine and sacrotuberous ligament (9 patients [4.8%]); Type III, entrapment in the Alcock canal on the medial surface of the obturator internus muscle (151 patients [79.9%]); and Type IV, entrapment at the distal branches of the pudendal nerve (25 patients [13%]).13

In the Type III category it is also possible to distinguish a Type IIIa with only obturator internus muscle involvement (49 patients [26%]) and a Type IIIb with involvement of both the obturator internus and piriformis muscles (102 patients [54%]). Specifically directed surgical releases of the pudendal nerve at the appropriate locations as well as specialized surgical steps depended on the subcategory.

The major role of obturator internus spasm in pudendal syndromes has been missed by most interested in this field. Although this role has been considered by some,26,41 there have been only 2 recent reports fully describing the management of this problem,13 one of which is a single patient case report.19

Treatment Outcomes

In patients with pudendal nerve distribution symptoms responsive to injections along the course of the pudendal nerve, targeted treatments proved to be highly effective. Long-standing relief in excess of 1 year with no known recurrence was achieved following injection alone in 24 patients (12%).

Fig. 6. Magnetic resonance neurography images obtained in a patient with pudendal entrapment just distal to the ischial spine. Axial neurographic view (A), with arrows indicating hyperintensity in the pudendal neurovascular bundle (PuNVB) on the medial aspect of the obturator internus muscle along the course of the Alcock Canal. Coronal neurographic (B), axial T1-weighted (C), and coronal T1-weighted (D) images showing cross-correlated anatomy (orange arrow and orange cross marks). Acetab = acetabulum; GM = Gluteus maximus; IS = ischial spine; IT = ischial tuberosity; IRF = ischio-rectal fossa; OI = obturator internus; PFM = pelvic floor muscles; SI = sacro-iliac joint.
A definite diagnosis of PNE was also made in 165 patients who did not obtain lasting relief from injection. Of these patients, 18 did not proceed to surgery, typically because they elected to undergo surgery elsewhere or simply did not want surgical treatment. Among the 147 surgical patients there were 185 operations (38 of which were second side surgeries completed during the same operating room visit or at a subsequent visit for bilateral entrapment). Only 7% of the 185 surgical cases were managed with simple distal pudendal neuroplasty (patients with Type IV syndrome not responsive to injections), with the remaining 93% requiring more extensive surgery.

Piriformis muscle resection was performed in 39% of the 200 patients in the study: those who had obturator internus and piriformis muscle involvement (Type IIIb, 75 operations) and in an additional 2% who had Type I syndrome (3 operations). This percentage represented the 2 groups of patients who had sciatic notch tenderness along with the pudendal symptoms.

Partial section of the sacrotuberous ligament was...
required in 12.5% of the surgeries (23 of 185 surgeries), including 10 cases with a Type II diagnosis (7 of these patients had surgery and 4 of the Type II surgeries were bilateral; each of the bilateral cases required sectioning of the ligament on both sides) and 13 with significant adhesion of the pudendal nerve to the sacrotuberous ligament, encountered as part of the inferior retrosciatic dissection.

Collectively, total therapeutic failure among surgically treated patients was uncommon and occurred in just over 5.5% of patients. Fair to moderate improvement—still > 2 or 3 out of 10 on the analog pain scale—occurred in an additional 13 patients (6.5%). Two patients (1%) experienced increased symptoms after treatment. The only significant surgical complication was a deep hematoma that developed 4 days after surgery in 1 patient and required surgical evacuation. The good to excellent outcome rate (improvement of ≥ 4 on the 10-point analog scale) was 68% at 3 months posttreatment and was sustained after 1 year of follow-up in 87% of those patients in whom 1-year data were available. The improved success rate at 1 year posttreatment relative to the rate at 3 months was attributable to the slow, steady improvement after 3 months; e.g., patients reported 0–3 points improvement at 3 months but > 4 points at 1 year.

Discussion

A pudendal nerve block at a location proximal to the point of entrapment that produces both numbness and pain relief in the involved area is the most convincing diagnostic finding to confirm the presence of pudendal neuralgia as the origin of symptoms. However, understanding that there are 4 major location categories for PNE syndromes—piriformis muscle/greater sciatic notch, ischial spine, Alcock canal/obturator internus muscle, and distal branches of the pudendal nerve—makes this principle highly effective.

A diagnostic block of the pudendal nerve proximal to its exit from the greater sciatic notch is difficult to accomplish, because the pudendal nerve is on the deep superior surface of the sciatic nerve as its superficial relation and adjacent to abdominal viscera as its deep relation. If a patient with possible proximal pudendal entrapment has sciatic notch tenderness, then injection of the piriformis muscle will often relieve the symptom. The introduction of Marcaine on the deep surface of the piriformis muscle is more likely to cause a sciatic block than a pudendal block.

A critical role of obturator internus muscle spasm in a large percentage of patients with pudendal neurological symptoms is another major finding in this study. Because of this role, in patients with tenderness to palpation on the medial aspect of the ischial tuberosity but nowhere else, an injection of Marcaine that relaxes the obturator internus muscle will typically provide abrupt and impressive relief of the pudendal symptoms with no associated block. This result makes a very convincing case for neuroplasty of the nerve to the obturator internus muscle as the critical step in relieving the PNE syndrome in these patients.

In patients with numbness or dysfunction rather than pain—particularly when due to a distal entrapment near the exit of the Alcock canal—specific local effectiveness of a steroid injection compared with the absent effect of a steroid injection at a distant location (for example, epidural or gluteus maximus) is useful although not definitively reliable.

In many cases, however, patients experiencing the symptom of numbness or urogenital dysfunction caused by obturator internus muscle spasm will report increased sensation and resolution of the dysfunction when Marcaine is introduced into the obturator internus muscle. The effects of muscle relaxation usually persist for ~ 24–48 hours, although a direct nerve block from Marcaine will typically resolve in 8–12 hours.

The potential to actually diagnose and relieve these syndromes on a reliable basis with small-scale, well-tolerated, minimal access surgical treatments should transform patient care. Even specialists familiar with diagnosing PNE still often advise patients that there is no known treatment, and this counsel is no longer correct.

Neurosurgeons are accustomed to treating severe spine, radicular, and CNS syndromes. However, pudendal syndromes can be totally disabling. Based on training, neurosurgeons tend to avoid seeing patients with urogenital, rectal, sexual, and perineal symptoms. Even for an experienced neurosurgeon, the identification and decompression of a small sensitive nerve such as the pudendal nerve is technically challenging, and this challenge is magnified for surgical specialties in which nerves are typically avoided. For this reason, it is appropriate that neurosurgeons with an interest in peripheral nerves become knowledgeable and comfortable with this type of entrapment syndrome. Although sexual and urogenital symptoms and end organs are involved, the diagnostic methodology and surgical issues are entirely appropriate for neurological and neurosurgical specialists.

Summary of Diagnostic Process

Patients with symptoms potentially referable to a pudendal nerve disorder can be sorted preliminarily into categories through physical examination: Type I, sciatic notch tenderness only; Type II, midischial tenderness; Type IIIa, obturator internus muscle tenderness only; Type IIIb, obturator internus and piriformis muscle tenderness; and Type IV, no palpable tenderness. Magnetic resonance neurography then can be used to identify unexpected causes, such as tumor, and to seek confirmatory abnormalities in nerve or adjacent structures (vessels or muscles). If the MR neurography is nondiagnostic, then patients should be carefully evaluated for myositis, autoimmune/rheumatological disorders, or unappreciated end-organ disorders. If that evaluation is nondiagnostic, then a ganglion impar block can be considered.

If the MR neurography findings are positive, then open MR image-guided injection is indicated for further diagnostic confirmation and possible percutaneous therapeutic effect: for Type I syndrome, inject the piriformis muscle; Type II syndrome, block the pudendal nerve at the ischial spine; Type IIIa syndrome, inject the obturator internus muscle; Type IIIb syndrome, inject both the piriformis and obturator internus muscles; and Type IV
syndrome, block the pudendal nerve in the area of the Alcock canal. If the injection produces no relief and no specific aggravation, proceed to immune/rheumatological evaluation and reassess end-organ causes; if these findings are negative consider a ganglion impar block. If a brief but definitive response is obtained (lasting 1 hour–3 days), consider performing surgery or administering a repeat injection of Botox for muscle and hyaluronidase for nerve in addition to the anesthetic agent and steroid. If a prolonged response is obtained (lasting 4 days to months), administer a repeat injection of Botox or hyaluronidase. If the second injection does not lead to a longer-lasting response, then surgery is recommended. If either the first or second injection produces lasting relief, no further treatment is needed.

Recommended surgical treatments for PNE syndrome are as follows: Type I, a piriformis muscle resection and neuroplasty of the pudendal nerve in the superior retrotrochiatric space (inside the sciatic notch) as well as in the proximal inferior retrotrochiatric space as the pudendal nerve exits the sciatic notch; Type II, neuroplasty of the pudendal nerve at the level of the ischial spine with possible sectioning of the sacrotuberous or sacrospinous ligaments, according to intraoperative findings; Type IIIa, neuroplasty of the nerve to the obturator internus muscle and the pudendal nerve in the inferior retrosciatic space, through the lesser sciatic notch, and the proximal Alcock canal; Type IIIb, same operation as that for Type I plus the procedure for Type IIIa; and Type IV, distal pudendal neuroplasty.

Conclusions

Understanding that the pudendal nerve can be entrapped at several different locations greatly improves the potential for success in treating patients whose symptoms affect the perineum and urogenital/anorectal systems indicate a potentially neurological basis. The availability of new methods to reliably diagnose and treat these entrapments will be helpful to a significant number of patients who have had little hope of relief in the past. Although neurosurgeons have traditionally avoided involvement with this class of pathology, it is clear that physicians in other specialties hesitate to undertake complex surgical treatments of nerves. The result has been an “orphan” condition with untreated patients who are often disabled by their symptoms. The diagnosis and treatment of pudendal nerve disorders should be incorporated into the knowledge base of neurosurgeons.

Disclosure

Dr. Filler is an owner of and holds stock in NeuroGrafix. He also holds a patent for MR Neurography.

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