Observational Study

Anatomical Variants of the Pudendal Nerve Observed during a Transgluteal Surgical Approach in a Population of Patients with Pudendal Neuralgia

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Background: Several studies have described the course and anatomical relations of the pudendal nerve. Several surgical nerve decompression techniques have been described, but only the transgluteal approach has been validated by a prospective randomized clinical trial. The purpose of this study was to describe the course of the nerve and its variants in a population of patients with pudendal neuralgia in order to guide the surgeon in the choice of surgical approach for pudendal nerve decompression.

Objectives: In order to support the choice of the transgluteal approach, used in our institution, we studied the exact topography, anatomical relations, and zones of entrapment of the pudendal nerve in a cohort of operated patients.

Study Design: Observational study.

Setting: University hospital.

Methods: One hundred patients underwent unilateral or bilateral nerve decompression performed by a single operator via a transgluteal approach. All patients satisfied the Nantes criteria for pudendal neuralgia. The operator meticulously recorded zones of entrapment, anatomical variants of the course of the nerve, and the appearance of the nerve in the operative report.

Results: One hundred patients and 145 nerves were operated consecutively. Compression of at least one segment of the pudendal nerve (infrapiriform foramen, ischial spine, and Alcock's canal) was observed in 95 patients. The zone of entrapment was situated at the ischial spine between the sacrospinous ligament (or ischial spine) and the sacrotuberous ligament in 74% of patients.

Anatomical variants were observed in 13 patients and 15 nerves. Seven patients presented an abnormal transligamentous course of the nerve (sacrotuberous or sacrospinous). A perineal branch of the fourth sacral nerve to the external anal sphincter was identified in 7 patients. In this population of patients with pudendal neuralgia, the pudendal nerve was stenotic in 27% of cases, associated with an extensive venous plexus that could make surgery more difficult in 25% of cases, and the nerve had an inflammatory appearance in 24% of cases.

Limitations: We obviously cannot be sure that the anatomical variants identified in this study can be extrapolated to the general population, as our study population was composed of patients experiencing perineal pain due to pudendal nerve entrapment and their pain could possibly be related to these anatomical variants, especially a transligamentous course of the pudendal nerve. The absence of other prospective randomized clinical trials evaluating other surgical approaches also prevents comparison of these results with those of other surgical approaches.

Conclusions: This is the first study to describe the surgical anatomy of the pudendal nerve in a population of patients with pudendal neuralgia. In more than 70% of cases, pudendal nerve entrapment was situated in the space between the sacrospinous ligament and the sacrotuberous ligament. Anatomical variants of the pudendal nerve were also observed in 13% of patients, sometimes with a transligamentous course of the nerve. In the light of these results, we believe that a transgluteal approach is the most suitable surgical approach for safe pudendal nerve decompression by allowing constant visual control of the nerve.

Key words: Surgical, operative technique, pudendal, neuralgia, transgluteal approach

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The pudendal nerve provides sensory and motor innervation to the perineum. It innervates the perineal muscles involved in anal and urinary continence and it also participates in sensory innervation of the perineum. It has a complex, initially intrapelvic course, which then becomes gluteal and then perineal.

The pudendal nerve is derived from the second, third, and fourth sacral ventral rami at the inferior part of the piriformis muscle. The nerve then passes through the greater sciatic foramen, in the infrapiriform foramen, and enters the gluteal region. The pudendal nerve then lies posteriorly to the ischial spine or sacrospinous ligament, medial to the internal pudendal vessels (1), and lateral to the inferior rectal nerve. After entering the perineum, the pudendal nerve travels in a fold of the obturator internus muscle fascia, Alcock's canal. The pudendal nerve gives rise to 3 main branches: inferior rectal nerve, perineal nerve, and dorsal nerve of the clitoris or penis. The inferior rectal nerve continues medially through the ischiorectal fossa to innervate the external anal sphincter and levator ani muscle.

Pudendal nerve entrapment can occur at various sites and can be responsible for persistent, disabling perineal pain (2,3). The diagnosis of pudendal neuralgia due to pudendal nerve entrapment is essentially clinical. No imaging modality is currently able to visualize pudendal nerve entrapment. Neurophysiological assessment mainly investigates motor function and normal neurophysiological results do not exclude pudendal nerve sensory neuropathy.

We have therefore previously described a set of criteria that are suggestive of the diagnosis of pudendal neuralgia (4,5). Several articles have described the course and anatomical relations of the pudendal nerve, but never in a population of patients with pudendal neuralgia. Several surgical nerve decompression techniques have been described; the transgluteal approach is preferred in our center, as it allows visualization of the entire extrapelvic course of the nerve (Fig. 1) (6). This approach is safe and provides encouraging results that have been validated by a prospective randomized clinical trial, in which 66% to 80% of patients improved. Transvaginal, transperineal, and laparoscopic approaches have been proposed, but none of them are able to visualize the entire course of the nerve or explore the main currently identified sites of entrapment. An entrapment syndrome is related to narrowing of the space surrounding the nerve, resulting in nerve compression and/or decreased nerve mobility.

The objective of this study was to describe the topography, anatomical relations, and anatomical variants of the pudendal nerve in a cohort of patients operated on for pudendal neuralgia via a transgluteal approach.

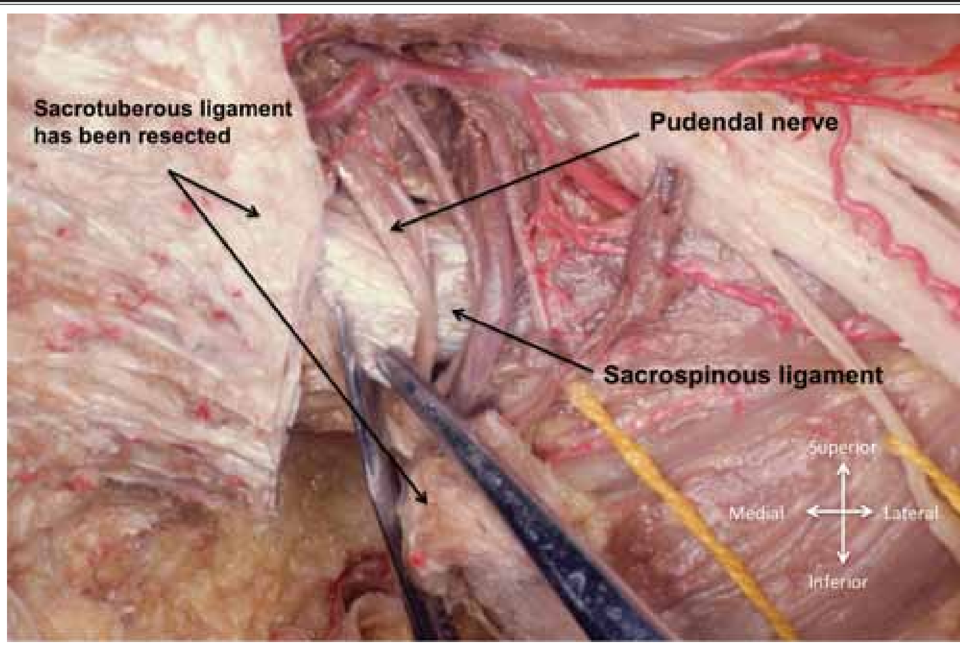

Fig. 1. *Anatomic view of the gluteal area after resection of the gluteus maximus muscle. The sacrotuberous ligament has been resected and the pudendal nerve lies over the sacrospinous ligament.*

Methods

One hundred patients were operated on via a transgluteal approach between January 2008 and December 2010. The operator was asked to record the topography, anatomical relations of the nerve, and all useful surgical findings in an operative report template that was validated prior to initiation of this study. Data collection was therefore retrospective. All patients presented with unilateral or bilateral pudendal neuralgia according to the Nantes criteria including an anaesthetic nerve block in the space between the sacrospinous and sacrotuberous ligaments (4).

Surgical findings were recorded in detail with particular attention to zones of entrapment, anatomical variants of the course of the nerve, and the morphology of the nerve (Fig. 2).

All patients gave their consent to this protocol for electronic data collection and use of data related to their clinical history.

Results

One hundred consecutive patients (145 nerves) were operated on. The cohort comprised 58 women and 42 men with a mean age of 56 years (18 – 79): 42% of patients were younger than 55 and 78% were younger than 70 years. Surgery was unilateral in 55 patients and bilateral in 45 patients.

Zone of Entrapment

Nerve entrapment was observed in at least one of the usual sites (infrapiriform foramen, interligamentous claw, and Alcock's canal [pudendal canal]) in 95 of the 100 patients (Table 1 and Fig. 3). The most common site of entrapment was at the level of the ischial spine between the sacrospinous ligament (or ischial spine) and the sacrotuberous ligament (74% of patients). A falciform process of the sacrotuberous ligament was demonstrated in 40% of cases, responsible for nerve compression as it enters Alcock's canal. Double entrapment involving both the infrapiriform foramen (E1) and the ischial spine (E2) was demonstrated in 6 patients (6 nerves). The most common combination was E2 and E3 (Alcock's canal) entrapment, observed in 33 patients (44 nerves). The E1 and E3 combination was observed in 8 patients (9 nerves) and only 4 patients presented entrapment at all 3 zones (E1, E2, E3). These zones of entrapment are characterized by impaired mobility of the nerve and the nerve can sometimes have a modified appearance in the zone of entrapment.

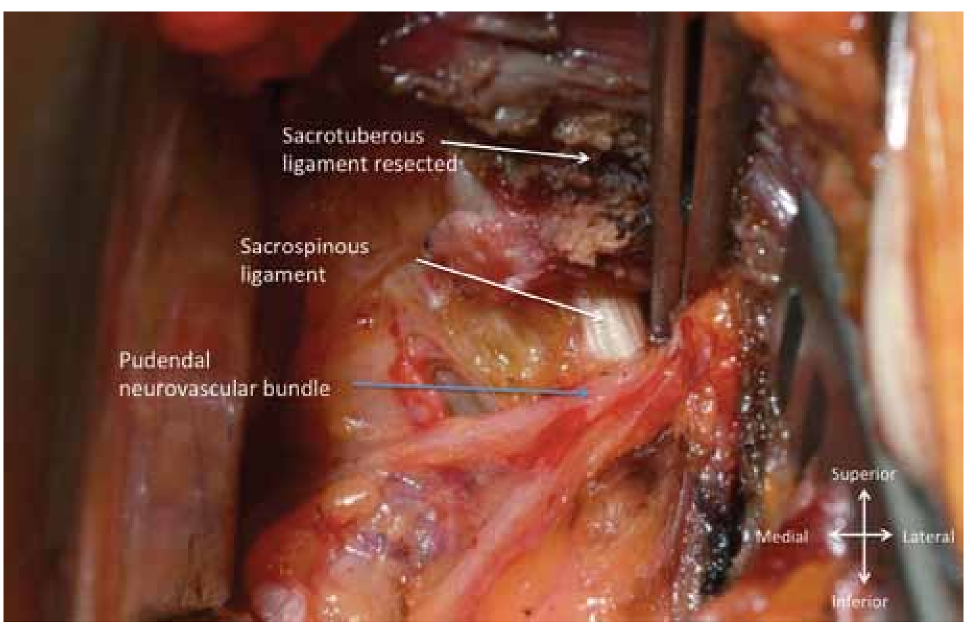

Fig. 2. *Surgical view after opening the gluteus maximus muscle (right side). The sacrotuberous ligament has been resected and the pudendal neurovascular bundle lies over the sacrospinous ligament.*

Anatomical Variants of the Course of the Pudendal Nerve

Anatomical variants were demonstrated in 13 patients (15 nerves). Seven patients (7 nerves) presented an abnormal transligamentous course of the nerve, 4 through the sacrotuberous ligament and 3 through the sacrospinous ligament (Table 2). A perineal branch of the fourth sacral nerve to the external anal sphincter was also identified in 7 patients and compression by the pudendal artery was observed for 2 nerves.

Anatomical Variants of the Ligaments

Anatomical variants were observed for both ligaments (Table 3), but more commonly concerned the sacrotuberous ligament, which was thicker than normal in 39% of cases (56 ligaments) and formed a fibrous sheath around the nerve in 8% of cases, making nerve dissection more difficult. Marked thickening of the sacrotuberous ligament can narrow the space between the sacrotuberous and sacrospinous ligaments, resulting in pudendal nerve entrapment.

Obturator internus muscle hypertrophy was observed in 7% of cases (10 muscles).

Table 1. *Sites of entrapment.*

	n	%
Infrapiriform foramen (E1)	15	10
Ischial spine (E2)	101	70
Alcock's canal (E3)	36	25
Falciform process	58	40

Appearance of the Nerve

In this population of patients with pudendal neuralgia, the pudendal nerve presented focal stenosis in 27% of cases (39 nerves); was associated with venous dilatations in 25% of cases (37 nerves); had an inflamed, fragile appearance, bleeding at the slightest contact, in 24% of cases (35 nerves) (Table 4); and had a normal appearance in 2% of cases (3 nerves).

Discussion

The objective of this study was to describe the intraoperative findings during transgluteal pudendal nerve decompression-transposition surgery. The main advantage of this approach is to allow constant visual control of the pudendal nerve, an essential requirement for any form of peripheral nerve surgery. A sin-

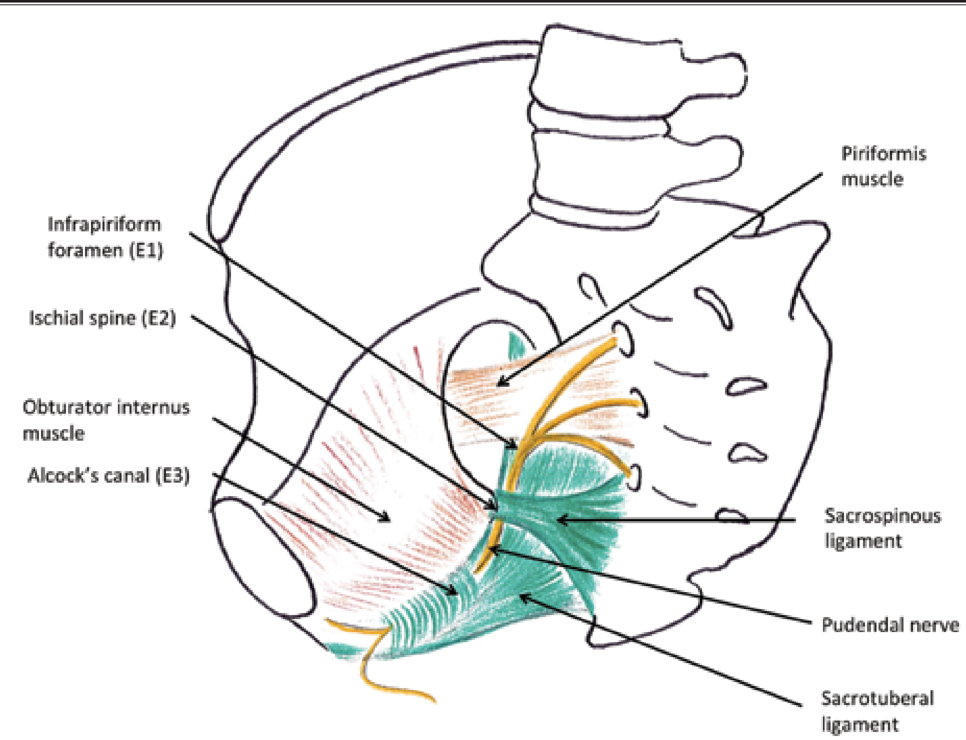

Fig. 3. Drawing showing the zones of entrapment (Table 1).

gle surgeon performed all surgical procedures and the operative findings for each nerve were clearly recorded in operating reports. This surgeon had previously performed a large number of cadaver anatomical studies (3). Each anatomical variant is significant and can potentially account for the patient's symptoms. No radiological examination is currently able to visualize the course of the nerve and identify potential zones of entrapment. The main objective of surgery is to restore nerve mobility, in the same way as in other better known nerve entrapment syndromes, such as carpal tunnel syndrome or ulnar nerve entrapment at the elbow. Moreover, in the 5 cases in which no zone of entrapment was identified (normal course and appearance of the nerve), surgery did not provide any significant improvement of pain.

After surgery, the surgical findings were explained to the patients, who eagerly awaited an explanation for the cause of their pain, often after many years of uncertainty and suffering.

A good knowledge of the course and organization of the extrapelvic pudendal nerve has a number of clinical implications, particularly for diagnostic anesthetic block of the pudendal nerve (7), which constitutes one of the Nantes criteria (4,5). Pudendal nerve decompression must be performed in the case of documented pudendal nerve entrapment (6).

Several authors have reported a number of anatomical variants of the pudendal nerve: variants affecting the origin of the nerve in the sacral plexus, and variants in terms of length, diameter, number of branches, and distribution (1,8-14). The present study focused on the surgical anatomy of pudendal nerve decompression, zones of entrapment, and the anatomical variants that must be recognized to allow safe pudendal nerve decompression. We therefore studied the course of the nerve from the infrapiriform foramen until the end of Alcock's canal.

These intraoperative anatomical findings have never been previously described in a population of patients with pudendal neuralgia. It is therefore not surprising to find a higher incidence of anatomical variants in this population compared to the general population.

The transgluteal approach appears to be the only surgical approach allowing confirmation of the diagnosis of pudendal nerve entrapment syndrome and visualization of potential zones of entrapment. This approach therefore allows surgical treatment based on anatomical arguments.

Table 2. *Abnormal course of the pudendal nerve.*

	n	%
Through the sacrotuberous ligament	4	3
Through the sacrospinous ligament	3	2
Perineal branch of the fourth sacral nerve	7	5

Table 3. *Variations of the sacrotuberous and sacrospinous ligaments.*

Sacrotuberous ligament	n	%
Thickening	56	39
Sheath around the nerve	12	8
Split	1	1
Thickening of the falciform process	6	4

Sacrospinous ligament	n	%
Thickening	3	2
Split	4	3
Ligament interspersed by muscle fibers	1	1

Table 4. *Appearance of the nerve.*

	n	%
Normal	3	2
Inflammatory	35	24
Stenosis	39	27
Fibrous sheath	39	27
Adipose transformation	19	13
Venous distension	37	25
Flattened appearance	4	3

The main zone of entrapment is situated at the ischial spine in the space between the sacrospinous ligament and the sacrotuberous ligament. The most traumatic structure is the more posterior sacrotuberous ligament, which compresses the nerve against the sacrospinous ligament (or ischial spine). This ligament must therefore be visualized, its morphology must be assessed, and it must be resected to permanently release the pudendal nerve. Section of the sacrospinous ligament then allows anterior transposition of the nerve, thereby decreasing the constraints placed on the nerve. Very often, the nerve is truly adherent to one of these ligaments. Dissection under visual control allows restoration of nerve mobility and perfectly safe resection of the ligaments, bearing in mind that section of these accessory ligaments of the sacroiliac joint has been shown not to induce any instability. Based on a

30-year experience with more than 5,000 cases, this approach is devoid of any major complications, especially sacroiliac joint instability (15).

Another important point is that, in some cases, branches of the nerve pass through these ligaments and can therefore only be dissected under direct visual control.

In more than 70% of cases, pudendal nerve entrapment was situated over the ischial spine in the space between the sacrospinous ligament and the sacrotuberous ligament (Table 1), as previously reported in the literature (3). Entrapment at several different sites was occasionally observed.

A transligamentous course of the nerve was demonstrated in 7% of patients and a perineal branch of the fourth sacral nerve was identified in 7% of patients (Table 2). These ligaments must therefore be constantly visible to avoid section of nerve branches passing through the ligaments. An anatomical study based on 15 female cadavers demonstrated that the pudendal nerve passed medially to the ischial spine and posteriorly to the sacrospinous ligament at a mean distance of 0.6 cm in 80% of subjects. In 20% of cases, the pudendal nerve was situated posteriorly to the ischial spine rather than posteriorly to the sacrospinous ligament (16). These authors also showed that the inferior rectal nerve crossed the sacrospinous ligament in 40% of subjects at an average of 1.9 cm medial to the ischial spine. A nerve to the levator ani muscle arose directly from the sacral nerves in 10 subjects (67% of cases). This nerve to the levator ani muscle was situated an average of 2.5 cm from the ischial spine (17,18). The authors described a “nerve-free zone” situated in the medial third of the sacrospinous ligament. These findings must be kept in mind to ensure safe dissection and section of the ligament. A recent article reported, as in our study, that the pudendal nerve may pass not only through the sacrospinous ligament, but also through the sacrotuberous ligament in 5% of cases (13). This anatomical variant of the course of the nerve can only be visualized via a transgluteal approach, which is the only surgical approach allowing decompression of this nerve.

Our results concerning anatomical variants of the course of the pudendal nerve are correlated with the data of the literature. In a large series of 37 cadavers (73 sides) studying divisions of the pudendal nerve over the sacrospinous ligament, 56.2% of pudendal nerves consisted of a single trunk, 31.5% were 2-trunked and 12.3% were 3-trunked (10). Fifteen inferior rectal nerves (20.5%) arose directly from the S4 nerve root, independently and never re-entered the pudendal nerve. Eight (11%) of 15 inferior rectal nerves crossed the sacrospinous ligament, therefore predisposing to nerve entrapment.

The present study also demonstrated anatomical variants of ligaments and obturator internus muscles (Table 3), which have not been previously reported in the literature. These anatomical variants are probably responsible for some types of pain; a hypertrophied obturator internus muscle can induce increased pressure on the inextensible fascia, thereby compressing the pudendal nerve.

This study describes, for the first time, the appearance of the nerve itself in this population of pudendal neuralgia patients (Table 4). Several different appearances of the nerve can be described. It often has an inflammatory and stenotic appearance, which can probably be attributed to the nerve entrapment.

The advantage of the transgluteal approach is that it allows constant visual control of the entire extrapelvic course of the pudendal nerve. This approach consequently facilitates pudendal nerve decompression by anticipating possible variants of the course of the nerve, which are observed in 13% of patients and 15% of nerves. The transgluteal approach is the only approach allowing access to the space between the sacrospinous and sacrotuberous ligaments, which is the main zone of pudendal nerve entrapment.

We believe that pudendal nerve decompression/transposition cannot be safely performed via a transperineal or transvaginal incision due to the poor visualization of the various zones of entrapment, a possible transligamentous course of the nerve (7% of patients), the course of the inferior rectal nerve, and the presence of a perineal branch of the fourth sacral nerve to the external anal sphincter, which must be preserved. The transgluteal approach is the only approach allowing anticipation of these variants to avoid accidental section of a branch of the pudendal nerve.

We obviously cannot be sure that the anatomical variants identified in this study can be extrapolated to the general population, as our study population was composed of patients experiencing perineal pain due to pudendal nerve entrapment and their pain could possibly be related to these anatomical variants, especially a transligamentous course of the pudendal nerve.

Our postoperative results have already been published and were identical in this cohort of patients (6), as 70% of patients were significantly improved, while 30% experienced persistent pain. These results are identical to those observed after herniated lumbar disc surgery.

Conclusion

In the absence of other studies evaluating other surgical approaches, these objective anatomical data of the gluteal region confirm that the transgluteal approach is the safest approach, as it allows constant visual control of the entire course of the nerve and recognition of the anatomical variants described. This approach also facilitates dissection, as the pudendal nerve is rapidly visualized after dissection of gluteus maximus and the sacrotuberous ligament.

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