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Decompression and Transposition of the Pudendal Nerve in Pudendal Neuralgia: A Randomized Controlled Trial and Long-Term Evaluation

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Abstract

Background: We assess that pudendal neuralgia is a tunnel syndrome due to a ligamentous entrapment of the pudendal nerve and have treated 400 patients surgically since 1987. We have had no major complication. We conducted a randomized controlled trial to evaluate our procedure.

Methods: A sequential, randomized controlled trial to compare decompression of the pudendal nerve with non-surgical treatment. Patients aged 18–70, had chronic, uni/bilateral perineal pain, positive temporary response to blocks at the ischial spine and in Alcock's canal. They were randomly assigned to surgery ($n = 16$) and control ($n = 16$) groups. Primary end point was improvement at 3 months following surgery or assignment to the non-surgery group. Secondary end points were improvement at 12 months and at 4 years following surgical intervention.

Results: A significantly higher proportion of the surgery group was improved at 3 months. On intention-to-treat analysis 50% of the surgery group reported improvement in pain at 3 months versus 6.2% of the non-surgery group ($p = .0155$); in the analysis by treatment protocol the figures were 57.1% versus 6.7% ($p = .0052$). At 12 months, 71.4% of the surgery group compared with 13.3% of the non-surgery group were improved, analyzing by treatment protocol ($p = .0025$). Only those randomized to surgery were evaluated at 4 years: 8 remained improved at 4 years. No complications were encountered.

Conclusions: In this study we demonstrate that decompression of the pudendal nerve is an effective and safe treatment for cases of chronic pudendal neuralgia that have been unresponsive to analgesia and nerve blocks. Following surgery, other medical interventions may be necessary.

Keywords: Neuralgia; Pelvic pain; Perineum; Sacral plexus; Neurosurgical procedure; Nerve compression syndromes; Prostatitis

1. Introduction

Patients suffering from chronic perineal pain may be assigned several diagnoses including prostatodynia, prostatitis, vulvodynia, chronic pelvic pain [1] syndrome, and levator ani syndrome. The diagnosis of pudendal neuralgia [2,3] is based on clinical symptoms: chronic debilitating perineal pain that is usually exacerbated in the seated position and relieved by standing, there is no nocturnal pain, normal imaging and the pain is unresponsive to usual analgesics. To our knowledge, there is no statistical study about the frequency of pudendal neuralgia in the literature.

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Our team treats 700 patients a year for perineal pain without any urologic, gynecologic and proctologic explanation. Among them, 200 have the diagnostic criterias of pudendal neuralgias as described above. 70 of them have no benefit with traditional antalgic treatment including the specific blocks. Finally, 40 patients out of 700 will be operated.

We have previously shown that the pudendal nerve can be subject to a ligamentous entrapment and that decompression and transposing the pudendal nerve may benefit patients with chronic pain [4]. We have operated on 400 patients since 1987 [5] and our surgical findings reinforce our view that a broad surgical release is necessary contrary to what has been proposed by others (and not evaluated) [6]. We wished to confirm objectively whether our clinical experience was valid. We therefore conducted a prospective randomized controlled trial to assess our procedure.

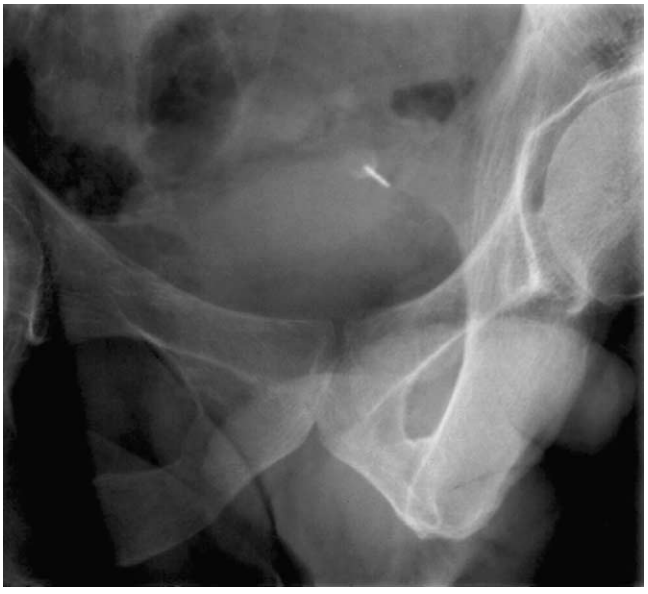

Fig. 1. Ischial spine block (fluoroscopy): Following antiseptic preparation of the skin, a 22G-90 mm spinal needle is inserted vertically just medially to the ischial spine. An injection is made with 4 ml of 1% lidocaine and 40 mg methylprednisolone acetate.

2. Methods

2.1. Study inclusion criteria

Patients eligible for inclusion in the study had chronic perineal pain of at least one year's duration in the area served by the pudendal nerve. Pain could be unilateral or bilateral, was exacerbated in the seated position, and was not marked at night. Patients had to be between the ages of 18–70, in good general health, have a pain intensity of at least 70 mm on a 100 mm visual analogue scale (VAS) [7] a minimum score of 3 on the behavior scale (Table 1), a positive diagnostic response to an anesthetic block of the pudendal nerve defined as numbness in the usually painful area after nerve block with a temporary reduction in pain while seated, persistence of perineal pain in spite of at least two steroid blocks of the pudendal nerve at the ischial spine and in Alcock's canal [8] no evidence from pelvic CT scans of pelvic or perineal pathology, and a depression score of 9 or below on the sub-scale of 6 items of the "depressive score" on the Hamilton depression rating scale [9,10]

2.2. Study design

The study was a sequential, randomized controlled trial, without blinding. Eligible patients gave informed consent after reading written information on the study. The ethics committee of the Nantes Centre Hospitalier et Universitaire (Comité Consultatif pour la Protection des Personnes en Recherche Biologique) approved the protocol. The source of funding was the CHU of Nantes. Patients who agreed to participate were assigned randomly to one of two treatment groups: surgery or control. Those in the surgery group were offered surgical decompression and transposition of the pudendal nerve. Medical treatment of the pain was identical in the two groups during follow up. It included anticonvulsant and antidepressant medications for neuropathic pain, relaxation and behavioral therapy. Steroid pudendal nerve blocks, were administered when the pain was unresponsive to analgesics. Several techniques have been described for carrying out pudendal nerve blocks. We used two principal transgluteal approaches: at the ischial spine (IS) (Fig. 1) and in Alcock's canal (Fig. 2). The diagnostic test is the temporary relief of the pain as a result of the local anaesthetic. A secondary therapeutic effect resulting from the steroid can be expected. In our previous experience, there is a reduction in pain in two thirds of patients with 2–3 blocks each, over a period of 6 months. We find it appropriate to perform the blocks at minimum intervals of 6–8 weeks and to limit the number of blocks to 2–4.

Physiotherapy was offered especially when a myofascial component was identified (triggers points localized in piriformis, obturator internis or levator ani muscles) and consisted of muscle stretches and relaxation [11].

The only difference, in the treatment, between the two groups is related to the fact that only the patients in the first group were operated on.

2.3. Surgical procedure

The transgluteal approach that we have described in 1989 [5] following Amarenco's hypothesis [12], allows all possible entrapments that we have detailed to be corrected in a single incision.

Table 1
Six-point behavior scale

(0)	No pain
(1)	Pain present but can easily be ignored
(2)	Pain present, cannot be ignored but does not interfere with daily activities
(3)	Pain present, cannot be ignored, interferes with concentration
(4)	Pain present, cannot be ignored, restricting all activities except basic needs such as washing and feeding
(5)	Pain present, cannot be ignored. Bed rest necessary.

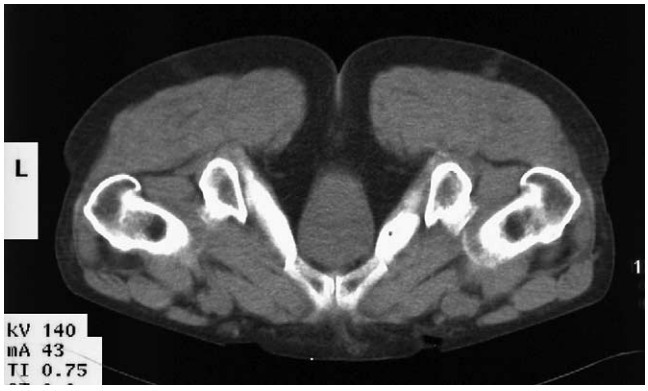

Fig. 2. Alcock's canal block (CT scan): The patient is installed in prone position. The ideal course of the needle is simulated on the screen by joining the posterior edge of the symphysis pubis to the internal edge of the ischium, passing between the obturator internus and its aponeurosis, and in extending this line back to the skin. The needle is inserted obliquely remaining in the vertical plane defining the section, parallel to the fascia of the obturator internus.

There are two main entrapment locations, eventually associated: in the claw between the sacrospinous and sacrotuberous ligaments near the ischial spine, in the Alcock's canal due to the falciform process of the sacrotuberous ligament and/or fascia of the obturator internus muscle.

Under general anesthesia, a gluteal incision of about 7 cm in length is made on both sides of a transversal line from the top of the coccyx, oriented obliquely according to the direction of the gluteus maximus muscle fibres, which are dissected and disinserted from the sacrotuberous ligament. The narrow section of the sacrotuberous ligament, located at the level of the ischial spine, is then resected transversally. The pudendal neurovascular bundle is then visible and released from the dorsal surface of the sacrospinous ligament. A simple retractor holding medially the ischio-rectal fat is sufficient to open the pudendal canal. It is then possible to perform a digitoclasic release of the nerve. If the fascia of the obturator is thickened or the falciform process is threatening, these can be incised. The sacrospinous ligament is cut and the nerve can then be transposed frontally to the ischial spine. One may then assess the diameter of the nerve, its shape (flattened or not), its inflammatory appearance, peritroncular fibrosis and satellite veins dilatations. The closure is effected in three planes. The sacrifice of the two ligaments has no morbid consequences for the sacro-iliac joint. The duration of surgery for one nerve is about 30 minutes. A 3–5 days hospital stay is required.

2.4. Outcome measures

The primary endpoint of the study was the proportion of patients judged to have improved at three months following surgery or after 3 months of medical treatments. Two principal outcome criteria were used: pain evaluated on a VAS, and quality of life, evaluated on a 6-point self-rated behavioral scale (Table 1). Treatment was considered effective if the pain score had decreased by at least 30 mm on the VAS and if the effect on quality of life rated less than 3 on the behavior scale. We followed up both groups at 1 year and the surgical group was assessed again at 4 years to find out whether improvement was maintained.

2.5. Statistical methods

Due to the difficulty in blinding surgical techniques, the study was not blinded. The Medical Computing and Statistics Unit carried out the randomization, balanced with unequal block sizes, independently of the investigators [13]. Efficacy in 60% of patients at 3 months was assumed in the surgical group and in 30% of patients in the non-surgery group. To establish a statistically significant difference of this size, with a p value of 5% in a one-tailed test and 90% power, using a sequential design, with analysis of efficacy every 8 cases and the triangular test, a maximum of 48 study participants was estimated.

3. Results

Between June 1994 and July 1996, 181 patients were seen in a multidisciplinary clinic for perineal pain and of these, 35 were eligible for the study (patients presenting with resistant pudendal neuralgia and potential candidates for surgical decompression). Three patients refused to participate. The other 32 (23 women and 9 men) were recruited in sequence and randomized, 16 to each of the study groups. The analysis of these 32 patients reached statistical significance and we there-

Table 2
Baseline characteristics of the two groups

	Surgery (n = 16) (mean ± S.D. (range))	Control (n = 16) (mean ± S.D. (range))	p value*
Gender	Female: 12 Male: 4	Female: 11 Male: 5	
Age	51.8 ± 11.8 (24–70)	56.4 ± 8.4 (40–67)	0.23
Duration of symptoms (years)	6.2 ± 4.8 (1–16)	3.4 ± 2.8 (1–30)	0.12
Mean pain score (VAS)	82.9 ± 8.7 (72–100)	85.2 ± 9.2 (72–100)	0.48
Mean behavior score	3.9 ± 0.7 (3–5)	4.1 ± 07 (3–5)	0.36
Hamilton score total	8.1 ± 4.6 (2–18)	11.2 ± 5.3 (5–22)	0.10
Hamilton score depression	3.6 ± 1.9 (0–8)	4.2 ± 2.4 (0–8)	0.50
Latency of bulbo-cavernous reflex right (ms)	51.0 ± 21.0 (30–86)	39.3 ± 5.0 (38–43)	0.71
Latency of bulbo-cavernous reflex left (ms)	43.1 ± 12.0 (32–65)	43.3 ± 7.4 (33–60)	0.17
Pudendal nerve latency right (ms)	4.2 ± 2.4 (1.7–7.8)	5.3 ± 0.6 (4.6–5.6)	0.29
Pudendal nerve latency right (ms)	4.6 ± 1.9 (0.6–7.7)	6.0 ± 2.6 (2.4–10)	0.16

* Non-parametric Mann–Whitney U test.

Table 3
Results at three months (primary end point)

	Intent to treat, *n* (%) (*n* = 32)		Per protocol, *n* (%) (*n* = 29)	
	Success	Failure	Success	Failure
Surgery	8 (50.0)	8 (50.0)	8 (57.1)	6 (42.9)
Control	1 (6.2)	15 (93.8)	1 (6.7)	14 (93.3)
Fisher exact test	0.0155		0.0052	

fore discontinued the study at this point, according to the sequential procedure decided, at its start. Table 2 shows that there were no significant differences between the groups with respect to baseline characteristics. Following allocation, 2 patients in the surgery group withdrew from the study before surgery, and one patient in the control group withdrew before the 3-month endpoint. These three cases are classed as failures in the intention-to-treat analysis at 3 months.

Table 3 shows that at 3 months, pain was alleviated significantly more often in the surgery than in the control group (intention-to-treat analysis): eight patients (50%) of the surgery group were improved versus 1 patient (6.2%) of the control group ($p = 0.0155$). In the analysis by treatment protocol the figures are 8 improved patients (57.1%) versus 1 patient (6.7%) ($p = 0.0052$). Among 15 patients of the control group, 6 had blocks during this period versus 4 out of 14 in the surgical group. At 3 months, 9 of the 16 patients in the control group wished to transfer to surgical treatment. They were not included in the surgical protocol and were all considered as failures of the non-surgical treatment at 12 months. Six others elected to continue with medical treatment.

At 12 months in the analysis by treatment protocol (Table 4) 10 patients in the surgery group (71.4%) and 2 controls (13.3%) had a successful outcome as defined for the study ($p = 0.0025$). Four of them are painless (VAS = 0). The surgery group was assessed again at 4 years. Eight of the 10 patients classified as successes at one year, remained so at 4 years. Seven of them had VAS pain scores of less than 15 mm and were considered completely cured. The eighth patient had a score of 38. Thus all patients considered as long lasting success had VAS scores under 40 at 12 months. The 4 patients who had surgery and were classified as failures at 12 months remained failures at 4 years. The 2 cases classified as a success at 12 months, but as failures at 4 years, had pain ratings of over 40 mm at the 12-month follow-up. It therefore appears that pain intensity at 1 year indicates long-term results whatever the initial score was.

In this study, we had no complication (infection, hematoma, neurological impairment, increasing of pain).

Table 4
Results at 12 months (secondary end point): analysis by treatment protocol (*n* = 29)

	Success, *n* (%)	Failure, *n* (%)
Success	10 (71,4%)	4 (28,6%)
Failure	2 (13.3%)	13 (86.7%)
Fisher exact test	0.0025	

4. Discussion

4.1. Overall results

This study demonstrates that surgical decompression of the pudendal nerve in intractable pudendal neuralgia is effective for patients in whom medical treatment, physical therapy and nerve blocks have failed. Although a success rate of 10 out of 14 at one year may not seem high, it is a good outcome for a chronic pain syndrome where more conservative treatments have failed. Patients included in this protocol had high VAS scores (7/10); 10 out of 14 clearly improved 3 points on the VAS by surgical treatment. Four of them are painless (VAS = 0). These results are quite different comparatively to the control group.

The surgical procedure is therefore validated and can reasonably be proposed for such patients. The efficacy of the surgery confirms the etiology of these postural perineal pain syndromes, previously considered as organs pathology (i.e., prostatodynias, vulvodynias, levator ani syndrom, ...).

Like in very common entrapment syndromes, no risk factors can be defined. Prevalence of women (60%) may be explain by parturition; sport as bike clearly exposes to this pathology [12].

4.2. Time-interval to evaluation

Assessment at 3 months was chosen for the primary end point and at 12 months for the secondary end point. This choice may seem inappropriate in the evaluation of chronic pain, but has been shown here to be relevant because although it does not necessarily give the definitive result, it nevertheless reveals the efficacy of the surgery compared with no surgical intervention. For ethical reasons, it seemed difficult to do the initial evaluation after more than 3 months. In fact, several patients of the control group decided to go out of the protocol in order to be operated. So, they have not been evaluated at 12 months. However we also followed up all patients at 12 months. It is interesting to note that

two additional patients were improved at 12 months compared with the 8 noted at 3 months. The interval of 12 months appears a suitable indicator of lasting success as patients having had a good result at 12 months retain this at 4 years if their pain score was below 40 mm at 12 months.

4.3. Design

The study was randomized but not blinded, as we believed blinding would not be ethically acceptable. We would have had to inflict a buttock incision as sham surgery for the control group. In any case, the surgical results far exceed what would be expected from any placebo effect. Placebo effects may be substantial when managing acute pain but have been shown to be much less so in chronic pain [14,15].

4.4. Safety and feasibility

No complications occurred during this series, confirming our previous experience of 400 patients that this procedure is safe and the operative risk is negligible.

Surgery never damages the nerve. As in classical surgery for entrapment syndroms, the goal is to release any source of entrapment, we never found any hypoesthesia nor motor complication after surgery.

4.5. Failure analysis

Several reasons can be suggested for surgical failures:

Diagnostic error is always possible although we offer surgery only to patients with a convincing clinical picture and we found entrapped nerves in all.

Long lasting or tight compression preoperatively could account for irreversible nerve fibers damage initiating chronic neuropathic pain. Relieving the entrapment might not be sufficient to initiate complete nerve recovery.

Incomplete release of the pudendal nerve could be responsible, particularly if the entrapment was at the distal part of the Alcock's canal and could not be reached through our surgical approach.

Postoperative fibrosis is also a recognized source of persistent pain following nerve decompression in general. However it is likely that patients with such fibrosis would experience a temporary improvement with secondary relapse rather than primary failure. Fibrosis may nevertheless explain the two cases in our series that had improved at 12 months but had relapsed at 4 years.

We could also have missed a contralateral entrapment in unilateral cases despite careful assessment. But, if so, we would expect the patient to experience postoperative pain mainly on the non-operated side.

Intractable chronic pain has significant emotional consequences and a psychological component is an integral part of the pain process. Although we tried to exclude patients with severe depression using a depression scale, the emotional, psychological and social distress may be important factors of resistance to treatment in all chronic pain syndromes.

5. Conclusions

This prospective, randomized study demonstrates that decompression and transposition of the pudendal nerve is a safe and effective treatment for patients with intractable pudendal neuralgia refractory to other treatments. This diagnosis must be clearly defined by clinical (chronic debilitating perineal pain, exacerbated in the seated position and relieved by standing, no nocturnal pain, normal imaging), neurophysiological, and diagnostic blocks tests. The success of a surgical approach also lends credence to our hypothesis that this type of pain is due to a tunnel syndrome and may offer a new approach to patients with chronic perineal, urogenital or anorectal pain previously misdiagnosed or abusively qualified as ''idiopathic'' or ''psychogenic''.

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References

[1] Wesselmann U, Burnett AL, Heinberg LJ. The urogenital and rectal pain syndromes. Pain 1997;73:269–94.
[2] Labat JJ, Robert R, Bensignor M, Buzelin JM. Les névralgies du nerf pudendal (honteux interne). Considérations anatomo-cliniques et perspectives thérapeutiques. J Urol Paris 1990;96:239–44.
[3] Turner ML, Marinoff SC. Pudendal neuralgia. Am J Obstet Gynecol 1991;165:1233–6.
[4] Robert R, Prat-Pradal D, Labat JJ, Bensignor M, Raoul S, Rebai R, Leborgne J. Anatomic basis of chronic perineal pain: role of the pudendal nerve. Surg Radiol Anat 1998;20:93–8.

[5] Robert R, Labat JJ, Lehur PA, Glemain P, Armstrong O, Le Borgne J, Barbin JY. Réflexions cliniques, neurophysiologiques et thérapeutiques à partir de données anatomiques sur le nerf pudendal(honteux interne) lors de certaines algies périnéales. Chirurgie 1989;115: 515–20.

[6] Shafik A. Pudendal canal syndrome as a cause of vulvodynia and its treatment by pudendal nerve decompression. Eur J Obstet Gynecol Reprod Biol 1998;80:215–20.

[7] Scott J, Huskisson EC. Graphic representation of pain. Pain 1976;2:175–84.

[8] Bensignor-Le Henaff M, Labat JJ, Robert T, Lajat Y. Douleur périnéale et souffrance des nerfs honteux internes. Cah Anesthesiol 1993;41:111–4.

[9] Hamilton M. Comparative value of rating scales. Br J Clin Pharmacol 1976;3:58–60.

[10] Bech P, Gram LF, Dein E, Jacobsen O, Vitger J, Bolwig TG. Quantitative rating of depressive states. Acta Psychiatr Scand 1975;51:161–70.

[11] Clemens JQ, Nadler RB, Schaeffer AJ, Belani J, Albaugh J, Bushman W. Biofeedback, pelvic floor re-education, and bladder training for male chronic pelvic pain syndrome. Urology 2000;56:951–5.

[12] Amarenco G, Lanoe Y, Ghnassia RT, Goudal H, Perrigot M. Syndrome du canal d'Acock et névralgie périnéale. Rev Neurol Paris 1988;144:523–6.

[13] Whitehead J. The design and analysis of sequential clinical trials. Chichester: Ellis Horwood; 1992, 298 pp.

[14] Hrobjartsson A, Gotzsche PC. Is the placebo powerless? An analysis of clinical trials comparing placebo with no treatment N Engl J Med 2001;344:1594–602.

[15] Price DD. Assessing placebo effects without placebo groups: an untapped possibility? Pain 2001;90:201–3.