Acta Chirurgica Belgica

ISSN: (Print) (Online) Journal homepage: www.tandfonline.com/journals/tacb20

Diagnosis and treatment of pudendal and inferior cluneal nerve entrapment syndrome: a narrative review

Katleen Jottard, Pierre Bonnet, Viviane Thill, Stephane Ploteau & Stefan de Wachter

To cite this article: Katleen Jottard, Pierre Bonnet, Viviane Thill, Stephane Ploteau & Stefan de Wachter (2022) Diagnosis and treatment of pudendal and inferior cluneal nerve entrapment syndrome: a narrative review, Acta Chirurgica Belgica, 122:6, 379-389, DOI: 10.1080/00015458.2022.2123138

To link to this article: https://doi.org/10.1080/00015458.2022.2123138

Published online: 16 Sep 2022.

ACTA CHIRURGICA BELGICA
2022, VOL. 122, NO. 6, 379–389
https://doi.org/10.1080/00015458.2022.2123138

Taylor & Francis
Taylor & Francis Group

REVIEW PAPER

Diagnosis and treatment of pudendal and inferior cluneal nerve entrapment syndrome: a narrative review

Katleen Jottard[a], Pierre Bonnet[b], Viviane Thill[a], Stephane Ploteau[c] and Stefan de Wachter[d]

[a]Department of Surgery, CHU Brugmann, Brussels, Belgium; [b]Department of Urology and Department of Anatomy, CHU Sart-Tilman, Liège, Belgium; [c]Department of Gynecology and Obstetrics, Center Hospitalier Universitaire, Nantes, France; [d]Department of UZA, Antwerp Surgical Training, Anatomy and Research Centre (ASTARC), Faculty of Medicine and Health Sciences, Wilrijk, Belgium

ABSTRACT

Aim: Pudendal and inferior cluneal nerve entrapment can cause a neuropathic pain syndrome in the sensitive areas innervated by these nerves. Diagnosis is challenging and patients often suffer several years before diagnosis is made. The purpose of the review was to inform healthcare workers about this disease and to provide a basis of anatomy and physiopathology, to inform about diagnostic tools and invasive or non-invasive treatment modalities and outcome.

Methods: A description of pudendal and inferior cluneal nerve anatomy is given. Physiopathology for entrapment is explained. Diagnostic criteria are described, and all non-invasive and invasive treatment options are discussed.

Results: The Nantes criteria offer a solid basis for diagnosing this rare condition. Treatment should be offered in a pluri-disciplinary setting and consists of avoidance of painful stimuli, physiotherapy, psychotherapy, pharmacological treatment led by tricyclic antidepressants and anticonvulsants. Nerve blocks are efficient at short term and serve mainly as a diagnostic tool. Pulsed radiofrequency (PRF) is described as a successful treatment option for pudendal neuralgia in patients non-responding to non-invasive treatment. If all other treatments fail, surgery can be offered. Different surgical procedures exist but only the open transgluteal approach has proven its efficacy compared to medical treatment. The minimal-invasive ENTRAMI technique offers the possibility to combine nerve release with pudendal neuromodulation.

Conclusions: Pudendal and inferior cluneal nerve entrapment syndrome are a challenge not only for diagnosis but also for treatment. Different non-invasive and invasive treatment options exist and should be offered in a pluri-disciplinary setting.

ARTICLE HISTORY
Received 10 May 2022
Accepted 4 September 2022

KEYWORDS
Pudendal neuralgia; pudendal entrapment; cluneal entrapment; chronic perineal pain

Introduction

Pudendal nerve (PN) and inferior cluneal nerve (ICN) entrapment can cause a neuropathic pain syndrome in one, many or all the sensitive areas innervated by these nerves [1,2]. Diagnosing this rare condition is challenging and patients often suffer several years before diagnosis is made. The purpose of the present review was to inform healthcare workers about this disease and to provide a basis for understanding and treating the condition. This review is mainly narrative, with its known limitations.

Anatomy and role of the pudendal nerve (Figure 1)

The PN has been referred to as the king of the perineum [3]. Indeed, the PN plays a major role in the fecal and urinary continence mechanisms and is important for normal sexual functioning. The PN has both motor and sensory functions and carries sympathetic fibers. It arises from the second, third, and fourth sacral ventral rami at the inferior edge of the piriformis muscle [4]. Before entering the gluteal region, the nerve passes through the infrapiriformis foramen, which is a part of the greater sciatic foramen. The nerve then passes posterior from the ischial spine or sacrospinous ligament (SSL), medial to the internal pudendal vessels, to finally enter the perineum through the Alcock's canal, a fold of the obturator internus muscle fascia. It continues to course through the pudendal canal (Alcock's canal), giving off three consecutive branches on its path: the inferior rectal (anal) nerve and its branches, the perineal nerve and its

CONTACT Katleen Jottard katleen.jottard@chu-brugmann.be Department of Surgery, CHU Brugmann, Brussels, Belgium

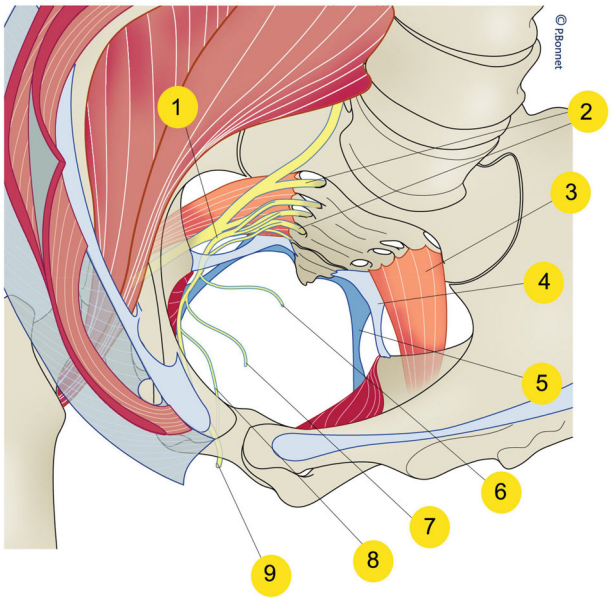

Figure 1. Pudendal nerve anatomy, anterior-cranial point of view. 1: pudendal nerve, 2: sacral nerve roots, 3: piriformis muscle, 4: sacrospinous ligament, 5: sacrotuberous ligament, 6: inferior rectal nerve, 7 + 8: perineal nerves, 9: dorsal nerve of the penis/clitoris (© P.Bonnet).

branches and the dorsal nerve of the penis or clitoris.

Anatomy and role of the inferior cluneal nerve (Figure 2)

The inferior cluneal nerve branches, which are purely sensitive, stem from the posterior femoral cutaneous nerve (PFCN), which is composed of fibers coming from the ventral branches of S1, S2 and S3 spinal nerves. The PFCN accompanies the sciatic nerve (SN), usually coursing medially or posteriorly to this nerve, while lying against the lateral aspect of the ischial tuberosity. The PFCN gives rise, medially, to the inferior cluneal nerves. They course below the inferior border of the gluteus maximus muscle and enter the skin of the buttocks. The number and course of the inferior cluneal nerves not only varies from one individual to another but also from one side to the other. *via* its numerous collateral branches, the posterior femoral cutaneous nerve innervates a very extensive area including the posterior surface of the thigh, the infragluteal fold, the skin over the ischial tuberosity, but also the lateral anal region, scrotum, or labium majus *via* its perineal branches. These perineal branches generally arise from the medial border of the posterior femoral cutaneous nerve, at the level of the inferior extremity of the ischial tuberosity and then courses subcutaneously over the origins of the hamstring muscles, transversely crossing over the tendons lateromedially [5,6].

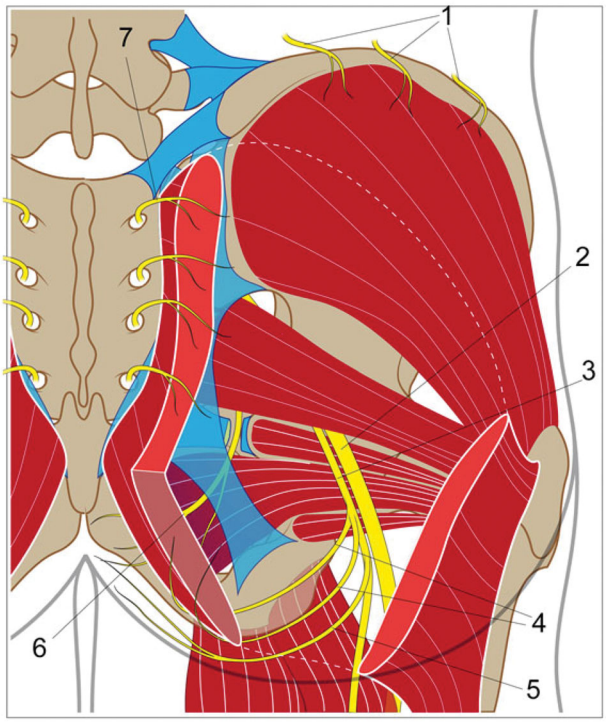

Figure 2. Nerves of the right-sided gluteal region, dorsal point of view. 1 = Superior cluneal nerves. 2 = Sciatic nerve. 3 = Posterior femoral cutaneous nerve. 4 = Inferior cluneal nerves. 5 = Perineal branches of posterior femoral cutaneous nerve. 6 = Pudendal nerve. 7 = Medial cluneal nerves. (© P.Bonnet).

Entrapment sites

For the PN several conflicting settings have been described during its passage: under the piriformis muscle, passing between the SSL and sacrotuberous (STL) ligament, entering the pudendal canal and passing the falciform process [7]. The entrapment site in the space between the SSL and STL is the most common, described in about 70% of the cases [8]. A transligamentous course of the PN through the SSL, which can even be calcified, has also been described. It is also at this level that the piriformis muscle can form a fibrous sheet around the nerve. After its passage between this ligamentous claw, the PN enters the pudendal canal. At the posterior border of the Alcock's canal, the PN passes over the falciform process of the STL, a fibrous sheet with a sharp upper border parallel to the medial side of the ischial bone. Finally, the pudendal vessels, which are often of considerable size and can be tortuous or dilated, can constrict the nerve.

The inferior cluneal nerve branches, including the perineal branch, running at the level of the inferior border of the ischial tuberosity, pass under a fibrous expansion, thicker on its ischial insertion than laterally, spread out between the ischial tuberosity and the deep gluteal fascia. This fibrous

Table 1. Diagnostic criteria for pudendal neuralgia by pudendal nerve entrapment [12].

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

expansion constitutes the principal site of entrapment of the nerve. The coexistence of adhesions between the nerve and the lateral aspect of the ischial tuberosity, can cause a lack of mobility of the nerve, which by itself, like in a tunnel syndrome, can lead to pain [6].

Clinical presentation

PN and/or cluneal nerve entrapment can cause a chronic neuropathic pain syndrome related to a loss of mobility of the nerves over their course which induces compression [1,2]. Pudendal and cluneal neuralgia coexist in 25% of patients [6]. The age of onset is in adult life, often without a clear etiology. The typical presentation of this pain syndrome is neuropathic pain, exacerbated in the sitting position in the sensitive areas previously described. Patients report a significant reduction or disappearance of pain during standing and in the decubitus position. Pain can be unilateral or bilateral. Because of the chronicity of this pain syndrome, patients often develop a peripheral and central sensitization due to an increase in the excitability of peripheral nerve fibers and the central nervous system so that normal inputs evoke exaggerated responses [9,10]. This is manifested in patients as allodynia or hyperalgesia. Furthermore, a pelvic hypersensitivity, partially related to muscular contractions, can enlarge the syndrome to a real pelviperineal pain syndrome causing urinary, sexual or defecation problems, making diagnosis even more challenging. Patients can also describe a feeling of rectal or vaginal foreign body.

Diagnosis

Diagnosing this rare condition, with unknown prevalence, is challenging and patients often suffer several years before diagnosis is made. A multidisciplinary team should be available to diagnose and treat these patients.

Physical examination

Physical examination in patients is often normal and is useful for excluding other causes of pelviperineal pain. Physical examination should exclude a perineal sensory deficit since this is highly suggestive of a sacral nerve root lesion. Presence of exquisite tenderness on palpation of the ischial spine during rectal or vaginal examination is not specific for pudendal entrapment syndrome [11]. Many anatomical structures are situated at this level, making this tenderness very difficult to interpret. Furthermore, it is also observed in asymptomatic subjects.

Complementary studies

Electrophysiological studies (electromyography and nerve conduction studies) only investigate large motor fibers and may not detect selective lesions of small sensory fibers. Other imaging, like Magnetic Resonance Imaging of the pelvic and lumbo-sacral region serve to exclude other causes of chronic pelviperineal pain. The pudendal and/or cluneal nerve block with local anesthesia is an important step in the diagnosis. A greater than 50% reduction in pain while sitting immediately after infiltration confirms the role of the pudendal and/or cluneal nerve. [12].

Nantes criteria (Table 1)

The Nantes criteria were discussed and validated by a multidisciplinary working party in Nantes, France on 23 and 24 September 2006 and then by members of the *Club d'électrophysiologie périnéale* (Francophone perineal electrophysiology club). These criteria were developed to define diagnostic criteria for pudendal and/or cluneal neuralgia by nerve entrapment [6,12]. In the absence of

pathognomonic imaging, laboratory and electrophysiology criteria, the diagnosis of pudendal neuralgia remains primarily clinical.

Treatment

Avoidance of painful stimuli

Avoidance of painful stimuli is an important step in the treatment. Patients should avoid prolonged sitting or activities as cycling or horse-riding. Custom-made seat cushions can be a solution for patients who need to sit for prolonged periods of time.

Physiotherapy

Pelvic muscle hypertonicity and myofascial trigger points can be present in patients. Manual therapy, dry needling and trigger point injections can be offered. Structural and biomechanical deviations like sacroiliac joint dysfunction, pelvic obliquities, lumbar spine pathology, leg length discrepancies and joint mobility should also be examined in all patients with pelviperineal pain.

Psychotherapy

Depression and anxiety are important contributors to the experience of pain and should therefore be taken into account in treating chronic pain patients [13]. Psychotherapy plays an important role in addressing the behavioral, cognitive, emotional, and social factors that both result from and contribute to pain-related dysfunction and distress. There are several distinct psychological interventions that differ in their theoretical approaches, therapeutic targets, and areas of efficacy [14].

Pharmacological treatment

Pharmacological treatment of chronic neuropathic pelvic pain is led by tricyclic antidepressants and anticonvulsants [15,16].

Pudendal and/or cluneal nerve block

Anesthetic nerve block of the pudendal and/or cluneal nerve serve mainly as a tool for confirming diagnosis since a greater than 50% reduction in pain while sitting immediately after infiltration confirms the diagnosis as described in the Nantes criteria [12]. Immediate improvement after pudendal infiltration, regardless of outcomes assessment, is achieved in 77–82% of patients, 3 months and 1 year post–procedure in 62% and in 6.8–12.2% respectively [15]. Adding corticosteroids to the nerve block does not seem to improve the outcome compared to local anesthetic alone [17].

Pulsed radiofrequency

Pulsed radiofrequency (PRF) is a successful treatment modality in patients with several neuropathic pain syndromes and it has also been described as a successful treatment option for pudendal neuralgia in patients non-responding to non-invasive treatment. A randomized controlled trial (RCT) published in 2018 compared PRF combined with PN block (PNB) using local anesthetics with PNB alone in 80 patients suffering from pudendal neuralgia according to the Nantes criteria [18]. At 3 months, they described a significant reduction of the visual analog scale (VAS) score from 5.7 ± 1.3 to 3.9 ± 2.1. The treatment effects were evaluated by the VAS, pain symptoms and local physical signs. The treatment effect was divided into four grades – 'completely cured,' 'significant positive effect,' 'effective' and 'invalid'. The 'completely cured,' 'significant effect' and 'effective' were marked as 'effective'. Three months after the procedure, the effective rate was 92.1% in the combined PRF group, but only 35.9% in the PNB group. No severe postoperative complications were described. Masala reported the results of 26 patients with pudendal neuralgia according to the Nantes criteria, who underwent CT-guided pulse-dose RF [19]. Outcome was only defined as changes in VAS. They described an improvement in all patients, also in the long term. In 2021, Withagen et al. published a case series of PRF for pudendal neuralgia with long-term follow up [20]. They evaluated quality of care by using the Patient Global Impression of Improvement scale (PGI-I), rating from 1 (very much better) to 7 (very much worse). The duration of pain relief after PRF varied between 6 weeks and 6 months and they repeated PRF when pain recurred. After 3 months, 79% of patients assessed their condition as (much) better (score 1 and 2). Over a long-term follow-up period, they reported a success rate of 89%. No major complications and no motor function loss occurred. Recently, Wang et al. reported the results of 70 patients with pudendal neuralgia according to the Nantes criteria, who underwent high-voltage long-duration PRF. Patients reported a significant pain relief, measured by decrease in VAS at each time point after treatment within a short-term follow period of 12 weeks. The therapeutic effect was

effective in 100% of patients with a recurrence rate of 11.4% at 12 weeks. No complications occurred [21].

Surgery (Table 2)

Pudendal and/or cluneal nerve decompression surgery is recommended after failure of medical treatment or minimally invasive percutaneous procedures such as pudendal/cluneal nerve block or pulsed radiofrequency, and it has proven its efficacy in the long term [22]. Different surgical approaches to liberate the nerve trunc in case of entrapment are described in the literature but only the open and endoscopic transgluteal approach give access to the PN at its entire course between the piriformis muscle up to the Alcock's canal and to the cluneal nerve and its potential entrapment site [22,23]. Only the open transgluteal approach proved its efficacy in the long term compared to medical treatment in a RCT [22]. Patients included had chronic pelviperineal pain for at least 1 year in the area served by the PN and met all five of the Nantes criteria for diagnosis of PN entrapment syndrome. Medical treatment was identical in the two groups (surgery or not) and consisted of anticonvulsant and antidepressant medication, relaxation, behavioral therapy, steroid PN blocks and physiotherapy. Surgery consisted of a transgluteal approach which necessitates a gluteal incision of approximately 5 cm. The STL was resected to have access to the pudendal neurovascular bundle. The falciform process was incised, if necessary, the pudendal canal was opened and the SSL was cut, after which the PN could be transposed anteriorly of the ischial spine. Sixteen patients were included in each study group. Outcomes were measured using the VAS and a quality-of-life scale, evaluated on a 6-point self-rated behavioral scale. At 12 months, 71.4% of the surgery group compared with 13.3% of the non-surgery group felt improvement. No complications occurred. The drawback of this open approach is the gluteal incision of 7–9 cm and the surgical trauma it therefore causes. The endoscopic transgluteal minimally invasive approach (ENTRAMI technique), described as a cadaver study in 2018, is based on the open transgluteal approach, but minimizes surgical trauma [23]. The postoperative outcome at 3, 6 and 12 months is similar as the outcome described in the open technique [24,25]. The 15 patients included met all 5 of the Nantes criteria for pudendal and/or cluneal nerve entrapment syndrome before surgery was proposed. The Numeric Pain Rating Scale (NPRS; range 0–10 with 0 = no pain and 10 = extreme pain/worst possible pain) for maximal pain intensity was recorded pre-surgery and 3-, 6- and 12-months post-surgery. Patient Global Impression of Change (PGIC, range 0–100%) was also assessed postoperatively. In case of bilateral surgery, patients were allowed to give five different scores, one for each side. Treatment failure was defined as PGIC $\leq$ 30%, good treatment response as PGIC $\geq$ 30%, and optimal response as PGIC $\geq$ 90%. The average duration of intervention (skin to skin) was 139 min (range 50–270 min) for bilateral pudendal and/or cluneal nerve liberation and 113 min (range 100–130 min) for unilateral pudendal and/or cluneal nerve liberation. No perioperative blood loss occurred. In 11 of 15 patients, only 2 trocars were necessary to complete the procedure. There were no immediate postoperative complications, besides a minor gluteal hematoma (grade 1 Clavien-Dindo classification), and 6 out of 15 patients were discharged on the first postoperative day. All other patients were discharged on the second postoperative day. Overall reduction of the average maximal NPRS score was from 9 (range 7–10) to 6 at 3 months (range 0–10; $p < 0.05$) and to 5 at 6 months (range 0–10; $p < 0.05$). At 3 months, 50% of patients had a good treatment response, increasing to 57% at 6 months. Optimal response (PGIC $\geq$ 90%) was found in 31% at 6 months. At 1 year after surgery, overall reduction of the average maximal NPRS score was from 9 (range, 7–10) at baseline to 5 (range, 0–10; *p*-value $<$.05). At 1 year 73% of patients declared to have a 'good treatment response' (PGIC $>$30%) and optimal treatment response (PGIC $\geq$90%) was found in 40% (*p*-value $<$.05), compared to 57% and 31% at 6-months respectively.

In the perineal pararectal approach described by Shafik, a vertical incision is made between the anus and the ischial tuberosity. The inferior rectal nerve serves as a landmark for identification of the PN at the level of the Alcock's canal, which is opened up to the ischial spine [26]. Eleven patients, diagnosed with idiopathic vulvodynia, were included in his study. They all had a positive response to a PNB and surgery was performed bilaterally. The vulvar pain improved gradually and disappeared in nine out of eleven patients. Perineal EMG and PN terminal motor latency (PNTML) scores were also improved in those nine patients. No postoperative complications occurred. Unfortunately, this approach only gives access to the Alcock's canal, which is only one of many sites of possible nerve entrapment.

Table 2. Summary table surgical procedures pudendal nerve release.

Name	Approach	Year	Type	nr	Criteria	Outcome	Advantage	Disadvantage
Shafik	Perineal	1998	Prospective	11	Vulvodynia	At 9 months: 9/11 patients improved	Reproducible	Blind procedure, only limited access, necessity of finger dissection
Bautrant	Trans ischio-anal	2003	Not clear	104	Pudendal neuralgia	86% of patients improved after 12 months	Endoscopic controle	Risk of hemorrhage and infection, and the necessity of finger dissection
Beco	Perineal	2004	Retrospective	74	PP pain, incontinence	11/26 patients pain improved	Reproducible	Blind procedure, only limited access, necessity of finger dissection
Robert	Transgluteal open	2005	RCT	32	Nantes criteria	At 12 months: 71.4 % improvement	Acces to PN entire course	Large gluteal incision
Possover	Laparoscopic	2009	Retrospective	18	Refractory ano-genital pain	Mean follow-up 21 months: decrease VAS score from 9.1 to 1.6 in 15 patients	Ability to diagnose endopelvic pathologies responsible for pelviperineal neuralgia	Invasive pelvic dissection
Erdogru	Istanbul technique	2014	Not clear	27	Pudendal nerve entrapment	At 1 month: VAS score dropped from 8.4 to 1.5	Omental flap protection of the nerve	Invasive pelvic dissection, long operating time
Beco	Endoscopic transperineal	2018	Prospective	113	Severe pudendal syndrome	50% pain reduction in 41.6% of patients in the long term	Better visualisation of PN	Only one operating hand
Bollens	Laparoscopic	202	Prospective	14	Exquisite tenderness on palpation of the pudendal nerve near the ischial spine during vaginal and rectal examination associated with anorectal, sexual and/or urinary symptoms	At 6 months: VAS score dropped from 6.8 to 2.2	Ability to diagnose endopelvic pathologies responsible for pelviperineal neuralgia	Invasive pelvic dissection
Jottard	Transgluteal endoscopic	2020	Prospective	15	Nantes criteria	At 12 months: 73 % improvement	Access to PN entire course and all entrapment sites minimal invasive	Learning curve

The same procedure was used by Beco et al. in his study in 2004 where 74 female patients were retrospectively analyzed [27]. Patients included suffered from pelviperineal pain or anal incontinence or urinary incontinence associated with at least two of the five following criteria:

a. Increased anal or perineal PNTML
b. Pathological EMG of the anal sphincter or bulbocavernosus muscles
c. Painful pudendal canal on rectal examination
d. Abnormal perineal sensibility
e. Painful skin rolling test.

Twenty-six of the included patients presented with pain. The authors describe 11 patients of those 26 reporting a disappearance of pain at 12 months. However, interpretation of these results is difficult since it is not clear where the pain was localized and if it was their main symptom if it was associated with incontinence and for how long patients were suffering. Furthermore, the inclusion criteria differ substantially from the criteria described in the Nantes criteria for patients suffering from PN entrapment syndrome. The combination of other procedures during nerve release surgery also makes it difficult to draw conclusions on the efficacy of PN liberation through this approach.

In 2007, Shafik reported a study to confirm the hypothesis that the PN could also be clamped between the SSL and STL. Treatment, consisting of SSL release through a perineal approach, was successful in 17 out of 21 patients with proctalgia which did not improve after his previous surgery [28].

In 2018, Beco et al. described an endoscopic transperineal pudendal decompression technique in which the perineal pararectal approach was combined with sectioning of the sacrospinous ligament and transposition of the PN [29]. The use of the pudendoscope made it possible to better visualize the anatomical structures, however, as stated by the authors themselves, the main disadvantage of operative pudendoscopy is the difficulty of operating with a single endoscopic 'dissecting hand'. Furthermore, to avoid vessel or nerve damage, it is mandatory to perfectly visualize all anatomical structures implicated. The study, in which different inclusion criteria than the Nantes criteria were used, found a 50% pain reduction in 41.6% of patients in the long term. However, it is unclear how many of those patients reported pelviperineal pain as their primary symptom.

The trans ischio-anal approach, transvaginal in women and transrectal in men, allows access to the infrapiriformis channel, the entire Alcock's canal and allows section of the SSL with endoscopic control [30]. A catheter for antalgic infusion is placed in the pudendal canal at the end of surgery. Bautrant reported the outcome of this approach in 104 patients who presented one major criterion plus two minor or major criteria.

The major criteria were the following:

1. Painful area in the PN path terminations
2. Reproduction of pain on pushing on the pudendal trunk (equivalent to the Tinel sign) and anatomical localization of the affected area
3. Successful injections of the anatomical site:
   - Lidocaine injection
   - Significant improvement or sedation of the pain for $>$ 12 hours.

The minor criteria were the following:

1. Neuropathic pain sensation
2. Existence of a painful position and/or antalgic (worse when sitting, better in decubitus)
3. Existence of an etiological factor or a trigger event
4. Lack of another cause of pain in the pelvic area.

Postoperatively three abscesses of the ischio-anal area and two hemorrhagic complications were described. After surgery 43 out of 104 patients (41%) reported an immediate pain disappearance measured by the VAS. At 1 year, 53 patients (of the only 62 patients reported on) declared an improvement. The drawback of this technique is the risk of hemorrhage and infection, and the necessity of finger dissection.

In 2004, Possover described the LANN-technique: laparoscopic neuronavigation to the pelvic nerves [31]: laparoscopic dissection and electric stimulation of the sacral roots guides to expose the PN.

No intraoperative complications nor postoperative functional morbidity occurred in the study reporting on this approach. However, the study lacks a precise step-by-step description of the technique, and the procedure has not been validated by a cadaver study which leaves the technique open for debate on the anatomical preciseness of structures identified. The feasibility of the

procedure on patients with pudendal pain was published in 2009 [32]. Eighteen patients with unilateral anogenital pain after sacrospinal fixation underwent the LANN-technique for PN release. The SSL was cut, and the PN was transposed. The procedure resulted in significant improvement in pain with a mean decrease in the preoperative VAS from 9.1 to 1.6 in 15 patients at a mean follow-up period of 21 months. The advantage of this technique is the ability to diagnose endopelvic pathologies responsible for pelviperineal neuralgia. However, as stated by the author himself, the access is less appropriate for pure PN dissection since it requires an invasive dissection.

The laparoscopic approach described by Erdogru et al., combines the laparoscopic transabdominal approach to section the SSL and for liberation of the PN in the Alcock's canal with the creation of an omental flap to allow protection of the PN (the Istanbul technique) [33].

The inclusion criteria of the 27 patients were:

A. Resistance of at least one of the three following symptoms to conservative treatments:
   i. Anogenital pain or perineodynia (on at least one side)
   ii. Painful lower urinary tract symptoms (LUTS)
   iii. Dyspareunia or painful erection–ejaculation

B. Association of at least two of the following criteria:
   i. Increased anal or perineal PN motor latency test (PNMLT)
   ii. Pathological EMG of the anal sphincter or bulbocavernosus muscles
   iii. Painful Alcock's canal and SSL on rectal or vaginal examination (on at least one side)
   iv. Decrease in pain with PNB.
   v. Painful skin-rolling test (Kibler fold test).

The VAS dropped from 8.4 to 1.5 at one month but it is unclear how many patients reported improvement and on which symptom pain release was reported (anogenital pain and/or painful LUTS and/or dyspareunia or painful erection or ejaculation). At 3 months, only 23 of the initial 27 patients were analyzed and 19 of them reported a more than 80% reduction in VAS. Postoperative complications were classified as Clavien-Dindo grade I or II and appeared in 19% of patients. As stated by the authors themselves, potential complications could include major vascular injury due to the closeness of the internal iliac vessels. The mean operating time was very long: between 155 and 300 min and advanced pelvic laparoscopic surgical experience is required when performing this procedure. Another drawback is, again, the extensive pelvic floor dissection necessary to access the PN.

Bollens et al. described the results of a laparoscopic approach in patients in whom an exquisite tenderness on palpation of the PN near the ischial spine during vaginal and rectal examination associated with anorectal, sexual and/or urinary symptoms was present [34]. The presence of pain in the area of the PN was not mandatory. Exclusion criteria comprised (1) patients who did not respond to PNB test using local anesthetics injection, (2) those who had exclusion criteria for the diagnosis of PN entrapment syndrome (purely coccygeal, gluteal, or hypogastric pain, exclusively paroxysmal pain, exclusive pruritus, and presence of imaging abnormalities able to explain the symptoms), and (3) those who had other organic pathological causes of pain. A step-by-step description of the procedure was reported to have access to the SSL for section, the pudendal neurovascular bundle for transposition as well as the proximal part of Alcock's canal for incision. Safety outcome measures for the procedure were reported for 235 patients. No serious Clavien-Dindo complications (III–V) were recorded. Finally, 32 patients filled in questionnaires before and after surgery. Three scores were used to assess the chief complaints. The Urinary Symptom Profile (USP) was used to evaluate urinary incontinence, overactive bladder and dysuria. The International Index of Erectile Function (IIEF-5) scores evaluated erectile function for male patients before and after the procedure. The Patient Assessment of Constipation Symptoms (PAC-SYM) was used to evaluate constipation. The intensity of perineal pain was estimated using VAS. Of those 32 patients, 14 patients reported pain in the area of the PN. With a mean follow-up period of 6 months, a decrease in VAS was seen from 6.8 to 2.2. However, it is unclear how many patients experienced improvement.

Pudendal neuromodulation

Pudendal neuromodulation (PNM) in case of chronic pelviperineal pain seems promising and different techniques have been described (perineal, transgluteal, laparoscopic, with or without neurophysiological guidance) [35–43].

Since optimal lead placement plays a key role in the maximum benefit of stimulation, improvements should be made regarding the correct

electrode placement. Despite using an optimized placement technique, it remains a blind procedure and the electrode cannot be fixed to prevent dislocation. Some complications, such as lead infection, pain, or lead migration, that are encountered in lead placement could be explained by these blind techniques. Furthermore, it is possible that suboptimal lead placement could partially explain the failure rates of neuromodulation.

In 2010, Carmel et al. published the results of pudendal neuromodulation in three patients suffering from pudendal neuralgia according to the Nantes criteria [44]. All three patients reported an improvement of 80% or higher, which was maintained for 2 years of follow-up. They used a pulse width of more than 450 μs which seemed to be an important stimulation parameter in these pain patients since a pulse width of less than 450 μs had an inferior outcome in one patient. Patients had a positive response to PNB to assess candidacy for peripheral nerve stimulation.

In a comparative pilot study of percutaneous pudendal implantation techniques, Heinze et al. showed a significant decrease of mean pain intensity during stimulation in patients with chronic pelvic pain: mean pain intensity decreased from 80 mm at baseline to 40 mm on a VAS scale after one month of stimulation. But inclusion criteria were not well described, and patients did not meet all the five of the Nantes criteria for PN entrapment syndrome. Peters et al. reported short-term 'improvement in pain' in all 19 subjects with pudendal neuralgia tested with pudendal neuromodulation [45].

In 2019, a cadaver study was published to implant a pudendal electrode using the previously described ENTRAMI approach [46]. The advantage is that the electrode is inserted under full visual control, hereby confirming the closeness of the electrode to the PN. Furthermore, the electrode can be fixed to the SSL to prevent dislocation. The disadvantage is the invasiveness of the procedure compared to percutaneous techniques. In 2020 the same authors published the early results of pudendal neuromodulation combined with PN release in case of chronic perineal pain syndrome using the ENTRAMI technique [47]. Patients eligible for inclusion had chronic perineal pain for at least 3 months in the area served by the PN and all met the five Nantes criteria. They combined PN release with short term pudendal neuromodulation using a PNE electrode.

At 1 month, the NPRS dropped from 9.5 at baseline to 3.5 ($p = 0.003$). Seventy-six percent of patients showed a global impression of change (PGIC) of $> 50\%$ at 1 month, and optimal treatment response (PGIC $\geq$ 90%) was found in 41% of patients.

Other

Other forms of neuromodulation have been described in the literature. Rigoard published a case report of a patient with chronic pelviperineal pain, who was unresponsive to a PNB and therefore not a candidate for release surgery [48]. The patient was implanted with a 16-contact surgical lead at the level of the conus medullaris, allowing multicolumn stimulation. Using transverse combinations, it was possible to obtain 100% paresthesia over the perineal area. Perineal and radicular pain was successfully relieved for up to 12 months.

A prospective study, published in 2015, proved the efficacy of spinal cord stimulation of the conus medullaris in 27 patients with refractory pudendal neuralgia after release surgery [49]. Of these patients, 74% were considered responders to treatment and 100% of implanted patients remained long-term responders.

Some articles also describe the use of dorsal root ganglion stimulation or sacral spine neuromodulation for patients with chronic pelviperineal pain syndrome. Unfortunately, due to the variety of pelviperineal pain syndromes included without clear distinction, conclusions about the efficacy of those therapies in case of pudendal neuralgia cannot be made [50,51]. The advantage of sacral neuromodulation is the test-phase prior to implantation. Nevertheless, neuromodulation for chronic pelviperineal pain syndromes remain off-label use.

Conclusion

Pudendal and cluneal nerve entrapment syndrome are a challenge not only for diagnosis but also for treatment. Different non-invasive and invasive treatment options exist, and these patients should ideally be treated in a pluri-disciplinary setting.

Future research should focus on clear patient inclusion criteria to compare treatment options.

Author contributions

Katleen Jottard: Conceptualization, Investigation, Writing – Original Draft Preparation, Writing – Review & Editing.

Pierre Bonnet: Writing – Review & Editing, Resources.

Viviane Thill: Writing – Review & Editing.

Stéphane Ploteau: Writing – Review & Editing.

Stefan de Wachter: Conceptualization, Writing – Review & Editing.

Disclosure statement

No potential conflict of interest was reported by the author(s).

References

[1] Hibner M, Desai N, Robertson LJ, et al. Pudendal neuralgia. J Minim Invasive Gynecol. 2010;17(2):148–153.

[2] Dellon AL. Pain with sitting related to injury of the posterior femoral cutaneous nerve. Microsurgery. 2015;35(6):463–468.

[3] Robert R, Labat JJ, Riant T, et al. [The pudendal nerve: clinical and therapeutic morphogenesis, anatomy, and physiopathology]. Neurochirurgie. 2009;55(4–5):463–469.

[4] Kinter KJ, Newton BW. Anatomy, abdomen and pelvis, pudendal nerve. Treasure Island (FL): StatPearls; 2021.

[5] Darnis B, Robert R, Labat JJ, et al. Perineal pain and inferior cluneal nerves: anatomy and surgery. Surg Radiol Anat. 2008;30(3):177–183.

[6] Ploteau S, Salaud C, Hamel A, et al. Entrapment of the posterior femoral cutaneous nerve and its inferior cluneal branches: anatomical basis of surgery for inferior cluneal neuralgia. Surg Radiol Anat. 2017;39(8):859–863.

[7] Robert R, Prat-Pradal D, Labat JJ, et al. Anatomic basis of chronic perineal pain: role of the pudendal nerve. Surg Radiol Anat. 1998;20(2):93–98.

[8] Ploteau S, Perrouin-Verbe MA, Labat JJ, et al. Anatomical variants of the pudendal nerve observed during a transgluteal surgical approach in a population of patients with pudendal neuralgia. Pain Phys. 2017;1(21;1):E137–E143.

[9] Levesque A, Riant T, Ploteau S, et al. Clinical criteria of Central sensitization in chronic pelvic and perineal pain (convergences PP criteria): elaboration of a clinical evaluation tool based on formal expert consensus. Pain Med. 2018;19(10):2009–2015.

[10] Gangadharan V, Kuner R. Pain hypersensitivity mechanisms at a glance. Dis Model Mech. 2013;6(4):889–895.

[11] Levesque A, Bautrant E, Quistrebert V, et al. Recommendations on the management of pudendal nerve entrapment syndrome: a formalised expert consensus. Eur J Pain. 2022;26:7–17.

[12] Labat JJ, Riant T, Robert R, et al. Diagnostic criteria for pudendal neuralgia by pudendal nerve entrapment (nantes criteria). Neurourol Urodyn. 2008;27(4):306–310.

[13] Woo AK. Depression and anxiety in pain. Rev Pain. 2010;4(1):8–12.

[14] Sturgeon JA. Psychological therapies for the management of chronic pain. Psychol Res Behav Manag. 2014;7:115–124.

[15] Tricard T, Munier P, Story F, et al. The drug-resistant pudendal neuralgia management: a systematic review. Neurourol Urodyn. 2019;38(1):13–21.

[16] Fall M, Baranowski AP, Fowler CJ, et al. EAU guidelines on chronic pelvic pain. Eur Urol. 2004;46(6):681–689.

[17] Labat JJ, Riant T, Lassaux A, et al. Adding corticosteroids to the pudendal nerve block for pudendal neuralgia: a randomised, double-blind, controlled trial. BJOG. 2017;124(2):251–260.

[18] Fang H, Zhang J, Yang Y, et al. Clinical effect and safety of pulsed radiofrequency treatment for pudendal neuralgia: a prospective, randomized controlled clinical trial. JPR. 2018;11:2367–2374.

[19] Masala S, Calabria E, Cuzzolino A, et al. CT-guided percutaneous pulse-dose radiofrequency for pudendal neuralgia. Cardiovasc Intervent Radiol. 2014;37(2):476–481.

[20] Krijnen EA, Schweitzer KJ, Wijck AJM, et al. Pulsed radiofrequency of pudendal nerve for treatment in patients with pudendal neuralgia. A case series with long-term follow-up. Pain Pract. 2021;21(6):703–707.

[21] Wang CL, Song T. The clinical efficacy of high-voltage long-duration pulsed radiofrequency treatment in pudendal neuralgia: a retrospective study. Neuromodulation. 2021;1–6.

[22] Robert R, Labat JJ, Bensignor M, et al. Decompression and transposition of the pudendal nerve in pudendal neuralgia: a randomized controlled trial and long-term evaluation. Eur Urol. 2005;47(3):403–408.

[23] Ploteau S, Robert R, Bruyninx L, et al. A new endoscopic minimal invasive approach for pudendal nerve and inferior cluneal nerve neurolysis: an anatomical study. Neurourol Urodyn. 2018;37(3):971–977.

[24] Jottard K, Bruyninx L, Bonnet P, et al. A minimally invasive, endoscopic transgluteal procedure for pudendal nerve and inferior cluneal nerve neurolysis in case of entrapment: 3- and 6-month results. The ENTRAMI technique for neurolysis. Int J Colorectal Dis. 2020;35(2):361–364.

[25] Jottard K, Bruyninx L, Bonnet P, et al. Endoscopic trans gluteal minimal-invasive approach for nerve liberation (ENTRAMI technique) in case of pudendal and/or cluneal neuralgia by entrapment: one-year follow-up. Neurourol Urodyn. 2020;39(7):2003–2007.

[26] 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(2):215–220.

[27] Beco J, Climov D, Bex M. Pudendal nerve decompression in perineology: a case series. BMC Surg. 2004;4:15.

[28] Shafik A, El Sibai O, Shafik IA, et al. Role of sacral ligament clamp in the pudendal neuropathy (pudendal canal syndrome): results of clamp release. Int Surg. 2007;92(1):54–59.

[29] Beco J, Seidel L, Albert A. Endoscopic transperineal pudendal nerve decompression: operative pudendoscopy. Surg Endosc. 2018;32(8):3720–3731.
[30] Bautrant E, de Bisschop E, Vaini-Elies V, et al. Modern algorithm for treating pudendal neuralgia: 212 cases and 104 decompressions. J Gynecol Obstet Biol Reprod (Paris). 2003;32(8 Pt 1):705–712.
[31] Possover M, Rhiem K, Chiantera V. The "Laparoscopic Neuro-Navigation" – LANN: from a functional cartography of the pelvic autonomous neurosystem to a new field of laparoscopic surgery. Minim Invasive Ther Allied Technol. 2004;13(5):362–367. Dec
[32] Possover M. Laparoscopic management of endopelvic etiologies of pudendal pain in 134 consecutive patients. J Urol. 2009;181(4):1732–1736.
[33] Erdogru T, Avci E, Akand M. Laparoscopic pudendal nerve decompression and transposition combined with omental flap protection of the nerve (Istanbul technique): technical description and feasibility analysis. Surg Endosc. 2014;28(3):925–932.
[34] Bollens R, Mjaess G, Sarkis J, et al. Laparoscopic transperitoneal pudendal nerve and artery release for pudendal entrapment syndrome. Surg Endosc. 2021;35(11):6031–6038.
[35] Heinze K, Hoermann R, Fritsch H, et al. Comparative pilot study of implantation techniques for pudendal neuromodulation: technical and clinical outcome in first 20 patients with chronic pelvic pain. World J Urol. 2015;33(2):289–294.
[36] Schmidt RA. Technique of pudendal nerve localization for block or stimulation. J Urol. 1989;142(6):1528–1531.
[37] Spinelli M, Malaguti S, Giardiello G, et al. A new minimally invasive procedure for pudendal nerve stimulation to treat neurogenic bladder: description of the method and preliminary data. Neurourol Urodyn. 2005;24(4):305–309.
[38] George AT, Dudding TC, Nicholls RJ, et al. A new minimally invasive technique for pudendal nerve stimulation. Colorectal Dis. 2012;14(1):98–103.
[39] Bock S, Folie P, Wolff K, et al. First experiences with pudendal nerve stimulation in fecal incontinence: a technical report. Tech Coloproctol. 2010;14(1):41–44.
[40] Possover M. A new technique of laparoscopic implantation of stimulation electrode to the pudendal nerve for treatment of refractory fecal incontinence and/or overactive bladder with urinary incontinence. J Minim Invasive Gynecol. 2014;21(5):729.
[41] Konschake M, Brenner E, Moriggl B, et al. New laparoscopic approach to the pudendal nerve for neuromodulation based on an anatomic study. Neurourol Urodyn. 2017;36(4):1069–1075.
[42] Possover M. A novel implantation technique for pudendal nerve stimulation for treatment of overactive bladder and urgency incontinence. J Minim Invasive Gynecol. 2014;21(5):888–892.
[43] Gregory NS, Terkawi AS, Prabhakar NK, et al. Peripheral nerve stimulation for pudendal neuralgia: a technical note. Pain Med. 2020;21(Suppl 1):S51–S55.
[44] Carmel M, Lebel M, Tu le M. Pudendal nerve neuromodulation with neurophysiology guidance: a potential treatment option for refractory chronic pelvi-perineal pain. Int Urogynecol J. 2010;21(5):613–616.
[45] Peters KM, Killinger KA, Jaeger C, et al. Pilot study exploring chronic pudendal neuromodulation as a treatment option for pain associated with pudendal neuralgia. Low Urin Tract Symptoms. 2015;7(3):138–142.
[46] Jottard K, Bonnet P, Bruyninx L, et al. The ENTRAMI technique: Endoscopic transgluteal minimal invasive technique for implantation of a pudendal electrode under full visual control: a cadaver study. Neurourol Urodyn. 2019;38(1):130–134.
[47] Jottard K, Bruyninx L, Bonnet P, et al. Pilot study: pudendal neuromodulation combined with pudendal nerve release in case of chronic perineal pain syndrome. The ENTRAMI technique: early results. Int Urogynecol J. 2021;32(10):2765–2770.
[48] Rigoard P, Delmotte A, Moles A, et al. Successful treatment of pudendal neuralgia with tricolumn spinal cord stimulation: case report. Neurosurgery. 2012;71(3):E757–E762; discussion E763.
[49] Buffenoir K, Rioult B, Hamel O, et al. Spinal cord stimulation of the conus medullaris for refractory pudendal neuralgia: a prospective study of 27 consecutive cases. Neurourol Urodyn. 2015;34(2):177–182.
[50] Mahran A, Baaklini G, Hassani D, et al. Sacral neuromodulation treating chronic pelvic pain: a Meta-analysis and systematic review of the literature. Int Urogynecol J. 2019;30(7):1023–1035.
[51] Hunter CW, Yang A. Dorsal root ganglion stimulation for chronic pelvic pain: a case series and technical report on a novel lead configuration. Neuromodulation. 2019;22(1):87–95.