Advances in the therapeutic approach of pudendal neuralgia: a systematic review

https://doi.org/10.1515/jom-2021-0119
Received April 20, 2021; accepted August 9, 2021; published online November 22, 2021

Abstract

Context: Although pudendal neuralgia (PN) has received growing interest over the last few years, diagnosis remains difficult, and many different therapeutic approaches can be considered.

Objectives: This article aims to provide an overview of the possible treatments of PN and investigate their efficacies.

Methods: Utilizing PubMed and ScienceDirect databases, a systematic review was carried out and allowed identification of studies involving patients with PN, as defined by Nantes criteria, and their associated treatments. Relevant data were manually reported.

Results: Twenty-eight articles were selected, totaling 1,013 patients (mean age, 49 years) and six different types of interventions. Clinical outcomes, most frequently quantified utilizing the Visual Analog Scale (VAS), vary greatly with both the therapy and time after intervention (from 100 to <10%). However, neither peri nor postoperative serious complications (grade > II of Clavien–Dindo classification) are reported. Although surgery seems to provide a higher proportion of long-term benefits, identifying the most efficient therapeutic approach is made impossible by the multitude of outcome measurements and follow-up frequencies. It should also be noted that literature is sparse regarding randomized controlled trials with long-term follow-up.

Conclusions: Although there are a number of modalities utilized for the treatment of PN, there are no current recommendations based on treatment efficacies. This seems to be largely in part caused by the lack of standardization in outcome quantification. Future research in this field should focus on prospective cohort studies with high levels of evidence, aimed at assessing the long-term, if not permanent, benefits of available therapies.

Keywords: pudendal neuralgia; systematic review; therapy; treatment.

Pudendal neuralgia (PN, also called Alcock canal syndrome, pudendal nerve compression, or pudendal nerve entrapment) emerges as a result of compression within the pudendal canal, also known as Alcock canal [1]. The pudendal canal is formed by the fascia of the obturator internus muscle. The first well-known symptom of this rare pathology is pain, mainly in the perineal region. Labat et al. [2] have summarized the different forms of pain originated by PN: burning, sensations of torsion or squeezing, electrical shocks of varying intensities, and/or sensitivity disorders. The disease is recognized by the French Social Security System and managed in its severe forms when severe deterioration of the living conditions of patients is reported [3]. The French Association for Pudendal Neuralgia (Association d’Information sur la Névralgie Pudendale et les Douleurs Pelvi-Périnéales) mentions that maintaining a professional activity is often compromised [4], and Beco, in a 2015 interview [5], mentions three patients who committed suicide due to pain.

Anatomy of the pudendal nerve

Arising from the second, third, and fourth sacral nerves, the pudendal nerve is an assembly of sensory, motor, and autonomic fibers. It follows a curved trajectory out of the pelvis through the greater sciatic foramen and between the piriformis and ischiococcygeus muscles. After a short gluteal path, the pudendal nerve spans dorsally around the sciatic spine, or generally around the distal insertion of the sacrospinous ligament [1, 6]. Its three different branches
are the rectal nerve (innervating the external anal sphincter), the dorsal nerve of the clitoris in females and of the penis in males, and the perineal nerve innervating the scrotum, labia majora, part of the muscles elevating the anus, the ischio and bulbocavernosus muscles as well as the bulb of the penis (Figure 1). Consequently, the role of the pudendal nerve is essential in the erection process and in both urinary and fecal continence [1].

A brief history of pudendal neuralgia

Zuelzer first described PN in 1915, but it was not until 1987 that Amarenco et al. [7] investigated perineal paralysis due to compression in Alcock’s canal in cyclists. However, there are several reasons why the prevalence of PN is rarely reported in the literature. First and foremost, health professionals lack training in treating PN, making diagnosis difficult. Second, the painful body area very often prevents patients from seeking medical attention: it is worth mentioning that “pudendal” comes from a Latin word meaning, “to be ashamed.” As a result, the International Pudendal Neuropathy Foundation estimates that the prevalence of PN may be around 1/100,000 of the world’s population (age: 50–70 years old; 2/3 females) [8]. In France, according to the Orphanet database [9], the prevalence of PN is estimated at 1/6,000.

No accurate etiology has been established yet. However, PN is often reported to arise from one or several of the following:

- Intensive practice of cycling or horseback riding (although less reported), leading to compression of the pudendal nerve
- Pelvic or orthopedic surgery
- Nerve stretching during childbirth
- Anomalies in the sacroiliac joint
- Prolonged sitting position, generally in professional activities

Several studies [10–13] enumerate these factors as triggers of PN. Cycling, and to a lesser extent horse riding [14], have been reported as the most common causes of PN. Both of these activities involve prolonged sitting with cyclic shocks, compressing the pudendal nerve at two possible locations: between the sacrotuberous and sacrospinous ligaments, or at the pudendal canal level (Figure 2).

It has been reported that 7–8% of cyclists suffer from PN with long-distance practice [15]. The main reason is that repeated impacts generate high perineal pressure, resulting in indirect compression of the pudendal nerve, thus increasing friction in Alcock’s canal [16]. Anatomically speaking, tissues are compressed between the saddle and

Figure 1: Innervation of the perineum showing the pudendal nerve (blue) and its ramifications: (A) dorsal nerve of the clitoris, (B) perineal nerve, and (C) inferior rectal nerve.
pubic symphysis, as well as between the saddle and ischial tuberosities [17].

Diagnosis of pudendal neuralgia

The diagnosis of PN is complex because it may easily be confused with gynecological disorders in females, or urological symptoms in males [18, 19]. Moreover, the very intimate location of this pathology often prevents patients from taking action. To date, the diagnosis of PN, as defined by Labat et al. [20] and commonly referred to as “Nantes criteria,” is still based on five clinical criteria:

- Pain in the pudendal nerve territory
- Increased pain while sitting
- Pain does not awaken patient from sleep
- Pain with no objective sensory impairment
- Pain relieved by anesthetic pudendal block

Once the first four criteria are met, a positive anesthetic pudendal block (fifth and final criterion) ascertains a diagnosis of PN. Other characteristic symptoms of neuropathic pain, such as alldynia, hyperpathia, or sympathalgia, may also appear.

Exclusion criteria include purely coccygeal, gluteal, pubic, or hypogastric pain, exclusively paroxysmal pain, painful defecation, and pruritus where objective sensitivity impairments observed by imaging may explain the pain [2]. In the end, positive local anesthetic blocks at the contact of the pudendal nerve remain the most efficient protocol in the diagnosis of PN [21].

Treatments

The therapeutic approach to PN is generally composed of four steps. Antiepileptics or antidepressants (e.g., pregabalin) are generally prescribed, followed by local muscle relaxers, such as opium or diazepam [8]. If ineffective, infiltrations coupled with local anesthetics and corticoids may be prescribed, targeting the distal insertion of the sacrospinous ligament in the sciatic notch and the fascia of the obturator internus in Alcock’s canal [22]. The effectiveness of this intervention has been reported in the literature [8, 23]. In the case of pelvic pain associated with hypertonia, physiotherapy is advised [8]. If all of the previously mentioned treatments prove ineffective, surgery may be considered as a last resort [24, 25], as long as PN has been present for more than 1 year [26, 27].
Finally, it is worth mentioning that few literature reviews are available regarding PN. To the best of the authors’ knowledge, only two systematic reviews have been published in this matter: one about drug-resistant PN by Tricard et al. [28] and the other on uncertain diagnoses of PN by Indraccolo et al. [29].

The review presented herein aims at analyzing the different therapeutic protocols tested and offered to patients suffering from PN and assessing their respective efficacies.

**Methods**

**Source**

In February 2021, EP and SM conducted the review that delivered an initial set of 1,345 articles published between 1949 and 2021. The main strategy adopted consisted of carrying out an exhaustive analysis of the scientific literature, based on the PubMed and ScienceDirect databases. The following keywords were employed to create a methodological search filter: “Alcock syndrome,” “Pudendal neuralgia,” “Pudendal nerve compression,” “Urogenital disorders,” and “Pudendal nerve entrapment.” A complementary strategy was aimed at obtaining additional information by examining and combining bibliographic references cited in the relevant articles obtained earlier. Other studies related to the topic of interest were therefore made available. In an attempt to increase the probability of finding further information, the table of contents listed in a number of medical journals as well as online documents from the AINP Association (French Association for the Information on Pudendal Neuralgia, approved by the Ministry of Health) were investigated.

EP, GP, F. Beaumont, F. Bogard, and SM reviewed the results of this initial search. Disagreements that were not resolved through discussion were arbitrated by MK.

**Inclusion criteria**

In this literature review, scientific papers and bibliographic studies about the various treatments of PN were selected. In view of the constant advances in medicine, only articles published after the year 2000 were included to ensure that results are correlated to current treatments. Although most articles are written in English and published in international journals, papers from French journals were also included to broaden the spectrum of knowledge in this rare disease.

**Exclusion criteria**

Papers mentioning perineal pain unrelated to PN were discarded. In addition, being that this is a literature review and not a study involving patient enrollment (randomized controlled, single or double blind, etc.) aimed at gathering statistically significant data, the exclusion criteria may match those selected in published statistical studies. However, being that PN is a rare disease, most works are case studies in which exclusion criteria are not explicitly defined.

**Fields of interest**

All selected articles were reviewed, and several sets of data were extracted. In a first step, the effectiveness of the investigated treatment, in terms of pain reduction, was recorded. The value generally consists of a Visual Analog Scale (VAS) score, even though several studies utilize different names for equivalent quantification. The second outcome of interest was the complications, as defined by the Clavien–Dindo classification [30]. This five-scale classification system aims at reporting the negative events after surgery in a simple and reproducible way, contrary to the subjective “minor” or “major” designations.

**Results**

Utilizing the criteria described in Section “Methods”, a set of 95 articles was built from PubMed and ScienceDirect.

Seven articles published prior to the year 2000 were discarded. Thirty-two other articles were found to not be PN-specific and subsequently discarded. Twenty-five articles only described the anatomy of the pudendal nerve or did not mention the efficacy of the treatment, so they were also discarded. Finally, three articles were not included because they reported procedures applied on animals or cadavers.

Once all of these filters and criteria were taken into account, a total of 28 articles were selected, encompassing 1,013 patients who were diagnosed with PN and treated. Figure 3 presents the synopsis of the selection protocol, which was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) [31].

Six different types of therapies are presented in the selected articles: transgluteal and laparoscopic decompressions, pudendal infiltration, radiofrequency, osteopathic manipulation, and nerve stimulator implantation. The most frequently utilized methods of objectification rely on the VAS, consisting in an 11-point value scale in which the score ranges from 0 (no pain) to 10 (unbearable pain), and a self-completed questionnaire on the patient’s quality of life (QoL, SF-36).

**General data of included studies**

Table 1 summarizes data about the selected articles, such as the authors’ names, the study population, the treatment type, the assessment method and its duration, and the remarkable results. Of all treatments adopted in the frame of PN, the most cited treatments are based on local infiltration targeted to the pudendal nerve and pudendal nerve decompression through surgery (including radiofrequency). A number of
Records identified from: ScienceDirect (n=1001) PubMed (n=344)

Records removed before screening: Duplicate records removed (n=51)

Records screened (n=1294)

Records excluded (n=1199)

Studies assessed for eligibility (n=95)

Reports excluded:
- Published before 2000 (n=7)
- Not PN-specific (n=32)
- No reported efficiency (n=25)
- Cadavers or animals (n=3)

Studies included in review (n=28)

**Figure 3:** Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flowchart.

**Table 1:** Main data of included studies.

| Study                      | Year | Journal                                      | Treatment                                      | No. of patients | Mean age, years | Female patients | Prospective study | Randomization | Control |
|----------------------------|------|----------------------------------------------|------------------------------------------------|-----------------|----------------|------------------|------------------|--------------|---------|
| Ramsden et al. [19]        | 2003 | American Journal of Physical Medicine and Rehabilitation | Transgluteal decompression                  | 1               | 25             | 0                | –                | –            | –       |
| Robert et al. [40]         | 2005 | European Urology                             | Transgluteal decompression                   | 32              | 54             | (23; 71.9%)      | Yes              | Yes          | Yes     |
| Filler [32]                | 2009 | Neurosurgical Focus                         | Transgluteal decompression                   | 147             | *              | *                | Yes              | –            | –       |
| Carmel et al. [43]         | 2010 | International Urogynecology Journal          | Electrode and generator implantation         | 3               | 50             | (3; 100%)        | –                | –            | –       |
| Hibner et al. [21]         | 2012 | The Journal of Minimally Invasive Gynecology | Transgluteal decompression                   | 10              | *              | (7; 70.0%)       | –                | –            | –       |
| Study                  | Year | Journal                                      | Treatment                        | No. of patients | Mean age, years | Female patients | Prospective study | Randomization | Control |
|-----------------------|------|----------------------------------------------|----------------------------------|----------------|----------------|-----------------|-------------------|---------------|---------|
| Lafave and Sutter [58] | 2012 | International Journal of Osteopathic Medicine | Osteopathic manipulation        | 1              | 31             | 0               | –                 | –             | –       |
| Vancailie et al. [46]  | 2012 | Pain Medicine                                | Pudendal infiltration           | 61             | 45             | (61; 100%)       | Yes               | –             | –       |
| Mamlouk et al. [51]    | 2014 | American Journal of Roentgenology           | Pudendal infiltration           | 31             | 55             | (28; 90.3%)     | –                 | –             | –       |
| Bendtsen et al. [47]   | 2016 | Regional Anesthesia and Pain Medicine        | Pudendal infiltration           | 3              | 52             | (2; –)          | –                 | –             | –       |
| Labat et al. [34]      | 2017 | BJOG International Journal of Obstetrics and Gynaecology | Pudendal infiltration | 201           | 57             | (122; 60.7%)    | Yes               | Yes          | Yes     |
| Petrov-K. et al. [45]  | 2017 | Pain Physician                               | Radiofrequency                  | 1              | 51             | (1; 100%)       | –                 | –             | –       |
| Fang et al. [35]       | 2018 | Journal of Pain Research                     | Radiofrequency                  | 77             | 45             | * Yes           | Yes               | Yes          | Yes     |
| Kastler et al. [23]    | 2018 | Pain Physician                               | Pudendal infiltration           | 95             | 52             | (61; –)         | –                 | –             | –       |
| Ricci et al. [52]      | 2018 | Brain Stimulation                            | Transcranial electrical stimulation | 1              | 76             | (1; 100%)       | –                 | –             | –       |
| Moscatiello et al. [36]| 2018 | Actas Urológicas Españolas                   | Laparoscopic decompression      | 1              | 60             | (1; –)          | –                 | –             | –       |
| Origo and Tarantino [59]| 2019 | Journal of Bodywork & Movement Therapies    | Osteopathic manipulation        | 1              | 40             | (1; 100%)       | –                 | –             | –       |
| Collard et al. [61]    | 2019 | Clinical Radiology                           | Radiofrequency                  | 10             | 60             | (8; 80.0%)      | –                 | –             | –       |
| Luther and Castellanos [37] | 2019 | Urology                                      | Transgluteal decompression      | 1              | 39             | 0               | –                 | –             | –       |
| Frank et al. [44]      | 2019 | Journal of Obstetrics and Gynaecology Canada | Radiofrequency                  | 7              | 44             | (7; 100%)       | –                 | –             | –       |
| Hunter and Yang [42]   | 2019 | Neumodulation                                | Dorsal root ganglion stimulation | 7              | 45             | (4; 57.1%)      | –                 | –             | –       |
| Kale et al. [48]       | 2019 | International Neurology Journal              | Pudendal infiltration           | 40             | 37             | (40; 100%)      | –                 | –             | –       |
| Ly et al. [49]         | 2019 | Pain Physician                               | Pudendal infiltration           | 91             | 54             | (62; 68.1%)     | –                 | –             | –       |
| Bollens et al. [63]    | 2020 | Surgical Endoscopy                           | Laparoscopic decompression      | 32             | 38             | (19; Yes)       | –                 | –             | –       |
| Gregory et al. [41]    | 2020 | Pain medicine                                | Stimulator implantation         | 1              | 76             | (1; 100%)       | –                 | –             | –       |
| Hodaj et al. [62]      | 2020 | Brain Stimulation                            | Radiofrequency                  | 18             | 60             | (13; 72.2%)     | –                 | –             | –       |
| Jottard et al. [38]    | 2020 | Neurology and Urodynamics                    | Transgluteal decompression      | 15             | *             | (8; Yes)        | –                 | –             | –       |
| Ramos et al. [39]      | 2020 | Central European Journal of Urology          | Transperitoneal neurolysis      | 1              | 23             | 0               | –                 | –             | –       |
| Dickson et al. [50]    | 2020 | ANZ Journal of Surgery                       | Pudendal infiltration           | 77             | 57             | (62; 80.5%)     | –                 | –             | –       |

* Indicates missing data.
complementary and/or novel treatments, such as the use of electrical stimulators or osteopathic manipulative treatment (OMT), have also been reported.

Outcomes

The outcomes reported by the selected articles are presented in Table 2. Even though most of them rely on VAS scores, the effectiveness indicators vary significantly from one study to another: treatment may be considered successful if VAS decreases by a given value or a given percentage. In some studies, the effectiveness threshold is not specified. Other studies utilize pain or QoL questionnaires to evaluate the treatment outcome, making quantification difficult. Case studies were not included in Table 2, since no treatment outcome assessment (in terms of percentage of the involved cohort) can be established.

Complications

As seen in Table 3, eight studies report complications. Two of them [21, 32] required surgery (grade III of the Clavien–Dindo classification [30]), and all others were ranked grade II or below.

Discussion

Overall level of scientific evidence

This review relies on analysis of 28 scientific papers matching the aforementioned inclusion criteria. Yet only three of them meet the standards of medical research regarding their level of scientific evidence, as advised by the French National Authority for Health [33], i.e., randomized controlled studies with adequate sample size: one on transgluteal surgery by Robert et al. applied to 32 patients, another by Labat et al. [34] on the combination of corticosteroids and local anesthetic with a cohort of 201 patients, and one last by Fang et al. [35] focusing on radiofrequency surgery with a population of 77 patients. In terms of population, 53% (15) of all studies have 10 or more patients, the remaining 47% (13) being single case studies (nine) or involving seven or fewer patients (four). Despite their intrinsic low level of evidence, studies involving fewer than 10 patients remain interesting to the extent that they may help identify trends or avenues of study, with a view to applying their treatments on a larger scale.

Outcome of available therapeutic approaches

Raw examination of the included studies shows that surgery, with no preferential site, seems to be the most efficient therapeutic approach: pain is significantly relieved [36–39], sometimes even completely [32, 40]. In all articles dedicated to this type of treatment, the patients’ QoL is reported as considerably improved, ensuring a return to optimal social life and overall health status. The most striking benefit of surgery is the duration of pain improvement, which varies from long-term (commonly 4 years) to permanent. Moreover, no serious postoperative complication was reported in any of the studies. In some cases, a second surgical intervention is required several months later to achieve complete recovery [21].

Many articles report patients treated by medication at the onset of pain in the pudendal nerve territory [34, 41–45], although the efficacy of the medication decreases after a few months [34]. Being that PN is a debilitating disease, prescribing antidepressants and anxiolytics makes it easier for patients to accept their pathology, but it has no effect on pain intensity. As mentioned by Tricard et al. [28], pharmacological treatments cannot be considered a sustainable solution for the treatment of PN.

Infiltration has a reported short-term efficacy [34, 46–50], while effects up to 1 year are observed in only a very small proportion of patients [51]. As expected in this type of treatment, it appears that infiltrations must be repeated to bring relief to patients, and their effect is only temporary. However, in cases of acute pain, it constitutes a good compromise due to much easier planning than surgery and the absence of recovery phase. Conversely, the effects of surgery (whether temporary or permanent) are perceived after about a year.

The use of electrical stimulation (transcranial [52], dorsal root ganglion [42]) has yielded satisfactory outcomes, although the reduced size of the cohorts as well as the lack of long-term follow-up make it difficult to evaluate the viability of this type of therapy. In addition, such invasive approaches cannot be used as a first line treatment.

In a 2018 study reporting a prospective, randomized controlled trial of 77 patients, Fang et al. [35] showed that Pulsed RadioFrequency (PRF) is able to significantly reduce pain after 3 months (the mean VAS score decreased by two). However, only the mean VAS score is reported, which explains why the study is not included in Table 2, because no success rate can be extracted. In addition, no follow-up is reported beyond 3 months, which makes it
Table 2: Outcome of treatments investigated in the selected studies.

| Study                | No. of patients | Effectiveness indicator method | Treatment                     | Immediate post-procedure success rate | Success rate after 1 month | Success rate after 3 months | Success rate after 6 months | Success rate after 1 year | Success rate after 2 years | Success rate after 4 years |
|----------------------|-----------------|--------------------------------|--------------------------------|----------------------------------------|---------------------------|------------------------------|----------------------------|--------------------------|----------------------------|-----------------------------|
| Hunter and Yang [42] | 7               | NRS decrease, no quantifiable indicator | Dorsal root ganglion stimulation | 100%                                  |                           |                             |                            |                          |                            |                             |
| Carmel et al. [43]   | 3               | VAS decreased by >60%           | Electrode and generator implantation | 80.0%                                 |                           |                             |                            |                          |                            |                             |
| Bollens et al. [63]  | 32              | VAS, USP, IIEF-5 and PAC-SYM   | Laparoscopic decompression        | 100%                                  |                           |                             |                            |                          |                            |                             |
| Bendtsen et al. [47] | 3               | VAS decrease, no quantifiable indicator | Pudendal infiltration            | 100%                                  |                           |                             |                            |                          |                            |                             |
| Mamlouk et al. [51]  | 31              | VAS decreased by >2            | Pudendal infiltration            | 6.8%                                  |                           |                             |                            |                          |                            |                             |
| Kale et al. [48]     | 40              | VAS decreased by >50%          | Pudendal infiltration            | 77.1%                                 |                           |                             |                            |                          |                            |                             |
| Vancaille et al. [46] | 61             | Complete loss of sensation reported in pain questionnaire | Pudendal infiltration            | 86.9%                                 |                           |                             |                            |                          |                            |                             |
| Dickson et al. [50]  | 77              | Immediate pain relief (excellent, good, some or delayed effect) | Pudendal infiltration            | 60.0%                                 |                           |                             |                            |                          |                            |                             |
| Ly et al. [49]       | 91              | VAS decreased by >50%          | Pudendal infiltration            | 29.5%                                 |                           |                             |                            |                          |                            |                             |
| Kastler et al. [23]  | 95              | VAS decreased by >50%          | Pudendal infiltration            | 63.2%                                 | 50.5%                     | 25.2%                       |                            |                          |                            |                             |
| Labat et al. [34]    | 201             | VAS decreased by >30%          | Pudendal infiltration            | 82.0%                                 | 13.4%                     |                            |                            |                          |                            |                             |
| Collard et al. [61]  | 10              | QoL improvement                | Radiofrequency                   | 60.0%                                 | 30.0%                     | 20.0%                       |                            |                          |                            |                             |
| Hodaj et al. [62]    | 18              | VAS decrease, no quantifiable indicator | Radiofrequency                   | 50.0%                                 |                           |                             |                            |                          |                            |                             |
| Frank et al. [44]    | 7               | VAS decrease, no quantifiable indicator | Radiofrequency                   | 100%                                  |                           |                             |                            |                          |                            |                             |
| Hibner et al. [21]   | 10              | VAS decreased by >2            | Transgluteal decompression        | 88.9%                                 |                           |                             |                            |                          |                            |                             |
| Jottard et al. [38]  | 15              | Average NPRS decrease, PGIC    | Transgluteal decompression        | 31.0%                                 | 40.0%                     |                             |                            |                          |                            |                             |
impossible to compare the efficacy of PRF to other non-invasive treatments. The more recent article by Frank et al. [44] on PRF mentions symptom relief between 6 weeks and 10 months, but the literature remains sparse regarding this technique, and no comparison on long-term benefits can be made with other therapeutic approaches.

As far as osteopathic medicine is concerned, several works report the effectiveness of OMT on peripheral nerve compression syndromes such as carpal tunnel syndrome [53–56]. In a 2012 report, the French National Institute of Health and Medical Research (INSERM) mentioned that the effectiveness of osteopathic medicine as a complementary treatment was moderate at best [57]. Nevertheless, it also stated that the literature was sparse and that the methodological limitations of the available studies were too significant for reliable conclusions to be drawn. In the present review, only two case studies—one by Lafave and Sutter [58] and the other by Origo and Tarantino [59]—were found regarding OMT-based approaches to PN. These two studies report a lasting effect on PN, with two different profiles: a male and a female, ages 31 and 40 respectively. It would seem that osteopathic medicine, as a first-intention manual therapy aimed at improving the overall motor function of the patient, can be complementary to surgery with a purpose of long-term, if not permanent, pain relief. However, both studies involve young subjects (<40 years old). Therefore, future research about the outcome of osteopathic medicine should therefore be based on older patients, given that PN mainly affects people between ages 50 and 70.

No article in the literature reports on studies about physiotherapy directly applied to PN, yet the therapy is a possible treatment for muscular hypertonia in PN patients [60]. Physiotherapy may be complementary to osteopathic medicine after a surgical intervention. New treatments that are less invasive than surgery have emerged over the last few years, such as PRF [35, 61, 62] or laparoscopic decompression [63], and proven their effectiveness in reducing pain and improving the patients’ QoL. However, the duration of these effects is limited to an average of 6 weeks.

In this review, the most conspicuous observation to emerge is that the follow-up of crucial variables, such as pain intensity and QoL, is ensured for durations that are far too diverse. In addition, the number of patients in most studies is too low, making the assertion of an optimal treatment of PN uncertain: studies involving larger patient cohorts are required to be able to generalize the effectiveness of a given treatment. The pathology was highlighted only recently, and it remains unaddressed or just lightly addressed in medical education. However, even though the amount of dedicated scientific papers remains very limited, it has been increasing during the last few years, which shows that there is growing

| Study                          | No. of patients | Manual therapy method                                      | Immediate post-procedure success rate | Success rate after 1 month | Success rate after 3 months | Success rate after 6 months | Success rate after 1 year | Success rate after 2 years | Success rate after 4 years |
|-------------------------------|----------------|-------------------------------------------------------------|---------------------------------------|----------------------------|-----------------------------|----------------------------|-----------------------------|-----------------------------|-----------------------------|
| Robert et al. [40]            | 32             | VAS decreased by ≥3                                         | 50%                                   | 71.4%                      | 68.0%                       | 87.0%                       | 81.0%                       | 87.0%                       | 75.0%                       |
| Filler et al. [32]            | 147            | Transgluteal decompression                                  | 50.0%                                 | 71.4%                      | 68.0%                       | 87.0%                       | 81.0%                       | 87.0%                       | 75.0%                       |

| Study                          | No. of patients | Manual therapy method                                      | Immediate post-procedure success rate | Success rate after 1 month | Success rate after 3 months | Success rate after 6 months | Success rate after 1 year | Success rate after 2 years | Success rate after 4 years |
|-------------------------------|----------------|-------------------------------------------------------------|---------------------------------------|----------------------------|-----------------------------|----------------------------|-----------------------------|-----------------------------|-----------------------------|
| BPI, Brief Pain Inventory questionnaire; C group, Control Group; CT, Computerized Tomography scan; E group, Experimental Group; EII-E, External Erector spinae; G, Group; HEDD, Hand-Eye-Dexterity; IEESI, Index of Erection Function; IIEF-5, International Index of Erectile Function; INH-CSI, International Society of General Urology–Chronic Prostatitis Symptom Index; I-P, International Prostate Questionnaire; IRT, Intrinsic Root Torque; M, Male; OAT, Ovarian Artery Transposition; PCOS, Polycystic Ovary Syndrome; PFS, Psychological Functioning Scale; PRF, Peri-Rectal Fat; R, Randomized Controlled; RCT, Randomized Controlled Trial; RC, Randomized Controlled Trial; RCCT, Randomized Controlled Trial; RCST, Randomized Controlled Trial; RCT, Randomized Controlled Trial; S, Serum; SF-36, Short-Form 36 Health Survey; SRI, Self-reported Improvement; TSK, Tampa Scale of Kinesiophobia; USP, Urinary Symptom Profile; VAS, Visual Analog Scale (0–10) for pain assessment. |
interest in PN. Hopefully, the related knowledge will be improved by utilizing studies with higher levels of scientific evidence, such as randomized controlled trials validated by reliable indicators such as the Physiotherapy Evidence Database (PEDro) scale [64]. More reliable conclusions should be able to highlight a relevant therapy.

### Comparative efficacies

As mentioned earlier, all nine single case studies [19, 36, 37, 39, 41, 45, 52, 58, 59], representing 32.1% of the studies selected after review, were not included in Table 2. The prospective, randomized control trial by Fang et al. [35]...
was not included because the efficacy was assessed utilizing a significant difference between the test and control groups. Among the remaining 19 (67.9%) cohort studies, the VAS (or equivalent) is utilized as an efficacy indicator in 15 (78.9%) studies.

Restricting the analysis to only those studies that rely on a significant numerical decrease of the VAS (or equivalent) score, i.e., VAS score decreased by >50% and/or by >3, it clearly appears that infiltration, regardless of the selected site, offers very satisfying immediate results. However, the beneficial effects disappear after 6 months. Radiofrequency-based approaches seem to provide similar results in terms of efficacy and duration, although further studies involving larger cohorts are needed to support this result. A very similar conclusion may be drawn regarding electrical stimulation, with very promising results reported even after 2 years. Minimally invasive procedures, such as laparoscopic decompression, exhibit an excellent outcome [63], but studies are lacking to support this observation. Finally, surgery in the form of transgluteal decompression shows very satisfying outcomes in the long term (up to 4 years).

Finally, it should be mentioned that osteopathic medicine may be utilized as a first-intention and/or complementary treatment, its efficacy being reported in other peripheral nerve compression syndromes. However, further studies are required, as only two single-case studies are detailed in this review.

Limitations

As discussed in the previous section, treatment may be considered successful in many different ways: six studies [38, 42, 44, 47, 62, 63] utilize mean VAS (or equivalent) score decreases, which makes it impossible to define an effectiveness indicator, while nine studies rely on different variations of the VAS score (e.g., relative variation, decrease by a given value). The remaining three studies utilize very different indicators, such as a complete loss of sensation [46], qualitative estimation of pain relief [50], or overall QoL improvement [61].

Consequently, and contrary to Nantes criteria that are now widespread in the diagnosis of PN, it can reasonably be concluded that there is no consensus regarding the best therapy assessment method and follow-up of patients. The multiplicity of both the result quantification items and the assessment criteria makes it difficult to analyze the clinical outcomes, and this calls for standardized indicators. Given the very recent advances in a number of therapeutic techniques of PN, emphasis should be placed on the establishment of international homogeneous criteria for patient assessment and follow-up. In such a way, all procedures could be compared directly.

Conclusions

In the present systematic review, 28 studies focusing on patients with PN and treated utilizing various approaches were investigated. It has been shown that to date, no actual scientifically robust consensus exists regarding either optimal treatment or follow-up. The scientific evidence level remains too low, due to a limited number of randomized controlled studies in the literature. It appears, however, that among all of the available treatments, surgery offers the best long-term results. Widespread knowledge of the clinical symptoms of PN should allow health professionals to directly diagnose this pathology, ensuring rapid care.

As far as osteopathic physicians are concerned, OMT can be offered to patients in addition to surgery of the pudendal nerve. However, the efficiency of such a combined approach remains to be scientifically validated by future research with a proven level of evidence.

Research funding: None reported.

Author contributions: E.P., M.K., and S.M. provided substantial contributions to conception and design, acquisition of data, or analysis and interpretation of data; E.P. and S.M. drafted the article or revised it critically for important intellectual content; G.P., F.B., and F.B. gave final approval of the version of the article to be published; and all authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Competing interests: None reported.

References

1. Robert R, Labat J-J, Riant T, Louppe J-M, Hamel O. The pudendal nerve: clinical and therapeutic morphogenesis, anatomy, and physiopathology. Neurochirurgie 2009;55:463–9.
2. Labat J-J, Delaviere D, Sibert L, Rigaud J. Approche symptomatique des douleurs pudendales chroniques. Prog Urol 2010;20:922–9.
3. La politique sanitaire encadrant la prise en charge des maladies névralgiques. Available from: https://www.senat.fr/questions/base/2009/qSEQ09121161.html [Accessed 1 Jun 2021].
4. Association d’Information sur la Névralgie Pudendale et les Douleurs Pelvi-Périnéales. Available from: https://www. association-aip.com/les-symptomes [Accessed 1 Jun 2021].
5. Fawcett K. Is this condition the secret source of your chronic pelvic pain? Available from: https://health.usnews.com/health-news/patient-advice/articles/2015/05/07/is-this-
condition-the-secret-source-of-your-chronic-pelvic-pain [Accessed 1 Jun 2021].
6. Maldonado PA, Chin K, Garcia AA, Corton MM. Anatomic variations of pudendal nerve within pelvis and pudendal canal: clinical applications. Am J Obstet Gynecol 2015;213: 727.e1–6.
7. Amarenco G, Lanoë Y, Perrigot M, Goudal H. A new canal syndrome: compression of the pudendal nerve in Alcock’s canal or perinal paralysis of cyclists. Presse Med 1987;16:399.
8. Hibner M, Desai N, Robertson LJ, Nour M. Pudendal neuralgia. J Minim Invasive Gynecol 2010;17:148–53.
9. Labat JJ. Le portail des maladies rares et des médicaments orphelins: la névralgie pudendale; 2014. Available from: https://www.orpha.net/consor/cgi-bin/Disease_Search.php?lng=FR&data_id=10804&Disease_Disease_Search_diseaseGroup=nevralgie-pudendale&Disease_Disease_Search_diseaseType=Pat&Maladie(s)/groupesdemaladies=Nevrailgie-pudendale&title=N%27Evralgie_pudendale&searc.
10. Pérez-López FR, Hita-Contreras F. Management of pudendal neuralgia. Climacteric 2014;17:654–6.
11. Amarenco G. Histoire de la névralgie pudendale: Une douleur presque sans… fondement. Pelvi-perineologie 2007;2:54–7.
12. Ziouziou I, Bennani H, Zizi M, Karmouni T, El Khader K, Koutani A, et al. Le syndrome du canal d’Alcock ou névralgie pudendale: un diagnostic à ne pas méconnaître. Can Urol Assoc J 2013;7:E486-9.
13. Ricchiuti VS, Haas CA, Seftel AD, Chelimsky T, Goldstein I. The vicious cycling: bicycling related pudendal nerve injury associated with avid bicycling. J Urol 1999;162:2099–100.
14. Murer S, Polidori G, Bogard F, Beaumont F, Polidori É, Kinne M. Perineal pressure in equestrians: application to pudendal neuralgia. Tavares J, Natal Jorge R, eds; ViPIMAGE 2019. Lecture Notes in Computational Vision and Biomechanics Springer; 2019, 34.
15. Asplund C, Barkdull T, Weiss BD. Genitourinary problems in women with chronic pelvic-perineal pain: a systematic review with a descriptive data synthesis. Neurourol Urodyn 2020;39: 890–7.
16. Shafik A. Pudendal canal syndrome. Description of a new syndrome and its treatment. Report of 7 cases. Coloproctology 1991;13:102–10.
17. Beco J, Cimov D, Bex M. Pudendal nerve decompression in perineology: a case series. BMC Surg 2004;4:15.
18. Ger GC, Wexner SD, Jorge JM, Lee E, Amaranth LA, Heymen S, et al. Evaluation and treatment of chronic intractable rectal pain– a frustrating endeavor. Dis Colon Rectum 1993;36:139–45.
19. Delion AL, Coady D, Harris D. Pelvic pain of pudendal nerve origin: surgical outcomes and learning curve lessons. J Reconstr Microsurg 2015;31:283–90.
20. Tricard T, Munier P, Story F, Lang H, Saussine C. The drug-resistant pudendal neuralgia management: a systematic review. Neurourol Urodyn 2019;38:33–21.
21. Indraccolo U, Nardulli R, Indraccolo SR. Estimate of the proportion of uncertain diagnoses of pudendal neuralgia in women with chronic pelvic-perineal pain: a systematic review with a descriptive data synthesis. Neurourol Urodyn 2020;39: 890–7.
22. Clavien PA, Barkun J, De Oliveira ML, Vauthey JN, Dindo D, Schuchli RD, et al. The Clavien-Dindo classification of surgical complications: five-year experience. Ann Surg 2009;250:187–96.
23. Shamseer L, Moher D, Clarke M, Ghersi D, Liberati A, Petticrew M, et al. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. BMJ 2015;349. https://doi.org/10.1136/bmj.g7647.
24. Filler AG. Diagnosis and treatment of pudendal nerve entrapment syndrome subtypes: imaging, injections, and minimal access surgery. Neurosurg Focus 2009;26:1–14.
25. Haute Autorité de Santé – Niveau de preuve et gradation des recommandations de bonne pratique; 2013. Available from: https://has-sante.fr/jcms/c_1600564/fr/niveau-de-preuve-et-gradation-des-recommandations-de-bonne-pratique-etat-des-lieux.
26. Labat JJ, Riant T, Lassaux A, Rioult B, Rabischong B, Khalfullah M, et al. Adding corticosteroids to the pudendal nerve block for pudendal neuralgia: a randomised, double-blind, controlled trial. BJOG An Int J Obstet Gynaecol 2017;124:251–60.
27. Fang H, Zhang J, Yang Y, Ye L, Wang X. Clinical effect and safety of pulsed radiofrequency treatment for pudendal neuralgia: a prospective, randomized controlled clinical trial. J Pain Res 2018; 11:2367–74.
28. Moscatiello P, Carracedo Calvo D, Yupanqui Guerra L, Rivera Martínez ME, de la Hoza A, Sánchez Encinas M. Robot-assisted pudendal neurolysis in the treatment of pudendal nerve entrapment syndrome. Actas Urol Esp 2018;42:344–9.
29. Luther RD 3rd, Castellanos ME. Successful treatment of penile numbness and erectile dysfunction resulting from pudendal nerve entrapment. Urology 2019;134:228–31.
30. Jottard K, Bruyninx L, Bonnet P, De Wachter S. Endoscopic transgluteal 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: 2003–7.
31. Ramos SA, Guimarães T, Bollens R. Urethral leak: an unusual symptom of pudendal nerve entrapment. Cent Eur J Urol 2020;73: 46–8.
32. Robert R, Labat JJ, Bensignor M, Glemain P, Deschamps C, Raoul S, et al. Decompression and transposition of the pudendal nerve.
in pudendal neuralgia: a randomized controlled trial and long-term evaluation. Eur Urol 2005;47:403–8.
41. Gregory NS, Terkawi AS, Prabhakar NK, Tran JV, Salmasi V, Hah JM. Peripheral nerve stimulation for pudendal neuralgia: a technical note. Pain Med 2020;21:551–5.
42. 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;22:87–95.
43. Carmel M, Lebel M, Tu LM. Pudendal nerve neuromodulation with neurophysiology guidance: a potential treatment option for refractory chronic pelvi-perineal pain. Int Urogynecol J 2010;21:613–6.
44. Frank CE, Flaxman T, Goddard Y, Chen I, Zhu C, Singh SS. The use of pulsed radiofrequency for the treatment of pudendal neuralgia: a case series. J Obstet Gynaecol Can 2019;41:1558–63.
45. Petrov-Kondratov V, Chhabra A, Jones S. Pulsed radiofrequency ablation of pudendal nerve for treatment of a case of refractory pelvic pain. Pain Med 2012;13:596–603.
46. Vancaille T, Eggermont J, Armstrong G, Jarvis S, Liu J, Beg N. Response to pudendal nerve block in women with pudendal neuralgia. Pain Med 2012;13:596–603.
47. Bendtsen TF, Parras T, Moriggl B, Chan V, Lundby L, Buntzen S, et al. Ultrasound-guided pudendal nerve block at the entrance of the pudendal (Alcock) canal: description of anatomy and clinical technique. Reg Anesth Pain Med 2016;41:140–5.
48. Kale A, Usta T, Basol G, Cam I, Yavuz M, Aytuluk HG. Comparison of ultrasound-guided transgluteal and finger-guided transvaginal pudendal nerve block techniques: which one is more effective? Int Neurourol J 2019;23:310–20.
49. Ly J, Scott K, Xi Y, Ashikyan O, Chhabra A. Role of 3 tesla mr neurography and CT-guided injections for pudendal neuralgia: analysis of pain response. Pain Physician 2019;22:E333–44.
50. Dickson E, Higgins P, Sehgal R, Gorissen K, Jones O, Cunningham C, et al. Role of nerve block as a diagnostic tool in pudendal nerve entrapment. ANZ J Surg 2019;89:695–9.
51. Mamlouk MD, vanSonnenberg E, Dekhkharghani S. CT-guided nerve block for pudendal neuralgia: diagnostic and therapeutic implications. Am J Roentgenol 2014;203:196–200.
52. Ricci R, Ghiggia A, La Rosa I, Milano A, Troni W, George MS, et al. Anodal Transcranial Direct Current Stimulation of the motor cortex reduces chronic pain in Alcock canal syndrome. Brain Stimul 2018;11:648–50.
53. Burnham T, Higgins DC, Burnham RS, Heath DM. Effectiveness of osteopathic manipulative treatment for carpal tunnel syndrome: a pilot project. J Am Osteopath Assoc 2015;115:138–48.
54. Siu G, Douglas Jaffe J, Rafique M, Weinik MM. Osteopathic manipulative medicine for carpal tunnel syndrome. J Am Osteopath Assoc 2012;112:127–39.
55. Sucher BM. Palpatory diagnosis and manipulative management of carpal tunnel syndrome. J Am Osteopath Assoc 1994;94:647–63.
56. Sucher BM, Hinrichts RN. Manipulative treatment of carpal tunnel syndrome: biomechanical and osteopathic intervention to increase the length of the transverse carpal ligament. J Am Osteopath Assoc 1998;98:679–86.
57. Barry C, Falissard B. Evaluation de l'efficacité de la pratique de l'ostéopathie. Inst Natl Santé Rech Med 2014;49:1–194.
58. Lafave M, Sutter B. Pudendal nerve entrapment in a bareback rodeo cowboy: a case study. Int J Osteopath Med 2012;15:78–82.
59. Origo D, Tarantino AG. Osteopathic manipulative treatment in pudendal neuralgia: a case report. J Bodyw Mov Ther 2019;23:247–250.
60. Cabanas-Valdés R, Calvo-Sanz J, Serra-Llobet P, Alcoba-Kait J, González-Rueda V, Rodríguez-Rubio PR. The effectiveness of massage therapy for improving sequelae in post-stroke survivors. A systematic review and meta-analysis. Int J Environ Res Publ Health 2021;18:4424.
61. Collard MD, Xi Y, Patel AA, Scott KM, Jones S, Chhabra A. Initial experience of CT-guided pulsed radiofrequency ablation of the pudendal nerve for chronic recalcitrant pelvic pain. Clin Radiol 2019;74:897.e17–23.
62. Hodaj H, Payen JF, Dumolard A, Delon-Martin C, Lefaucheur JP. Treatment of pudendal neuralgia by high-frequency rTMS of the medial wall of motor cortex bilaterally using an angled figure-of-eight coil. Brain Stimul 2020;13:1412–3.
63. Bollens R, Mjaess G, Sarkis J, et al. Laparoscopic transperitoneal pudendal nerve and artery release for pudendal entrapment syndrome. Surg Endosc 2020. https://doi.org/10.1007/s00464-020-08092-4.
64. Blobaum P. Physiotherapy evidence database (PEDro). J Med Libr Assoc 2006;94:477–78.