

Proctalgia and pudendal nerve entrapment: an association to know

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ABSTRACT

Introduction: Pudendal nerve entrapment (PNE) is a clinical syndrome, little known in the field of General Surgery, which includes a wide range of urinary, sexual and proctological symptoms. The interest for general surgeons lies in the whole clinical study that these patients may present as regards proctology. Complex diagnosis requires a sequential treatment that includes different tools.

The aim of this study is to present PNE from the point of view of general surgery by showing a study carried out in patients with proctalgia to assess the results at follow-up after 6 months.

Methods: We present an observational study evaluating 53 proctalgia patients in a PNE context who have undergone hydrocortisone puncture of the pudendal nerve, for anesthetic reasons, and its terminal branches, observing its immediate response in clinical terms of pain and in follow-up as from six months.

Results: Based on the data obtained from our sample, it may be seen that the treatment with these punctures improves 79.25% of patients in the period immediately after puncture and 39.62% of patients maintain this improvement after the six months.

Conclusions: We conclude that in the presence of proctalgia, surgeons should ignore the presence of PNE and that, according to our study, corticosteroid puncture treatment for anesthesia is an effective treatment option that provides relief to a significant percentage of patients.

Key words: Proctalgia, pudendal nerve entrapment, pudendal neuropathy, pudendal nerve.

RESUMEN

Introducción: El síndrome de atrapamiento del nervio pudendo (SANP) es una entidad clínica, poco conocida en el ámbito de la Cirugía General, que comprende un amplio abanico de síntomas urinarios, sexuales y proctológicos. El interés para el cirujano general radica en toda la clínica que pueden presentar estos pacientes en la esfera proctológica. De diagnóstico complejo, exige un tratamiento secuencial que incluye distintas herramientas. El objetivo del presente estudio es exponer el SANP desde el punto de vista de la cirugía general, exponiendo un estudio realizado en pacientes afectados de proctalgia para valorar los resultados en el seguimiento a partir de los seis meses.

Métodos: Presentamos un estudio observacional que evalúa 53 pacientes afectados de proctalgia en el contexto de un SANP que han sido sometidos a tratamiento con punción-hidrodistensión corticoideo-anestésica del nervio pudendo y sus ramas terminales, observando su respuesta inmediata en términos clínicos de dolor y en seguimiento a partir de los seis meses.

Resultados: De los datos obtenidos de nuestra muestra, se observa que el tratamiento con dichas punciones mejora al 79,25 % de los pacientes en el periodo inmediato tras la punción, y el 39,62 % de los pacientes mantienen dicha mejoría a partir de los seis meses.

Conclusiones: Concluimos que ante la presencia de proctalgia, el cirujano debe descartar la existencia de un SANP y que, según nuestro estudio, el tratamiento con punción corticoideo-anestésica es una opción eficaz de tratamiento que logra mejorar a un importante porcentaje de pacientes.

Palabras clave: Proctalgia, atrapamiento del nervio pudendo, neuropatía pudenda, nervio pudendo.

INTRODUCTION

PNE is a syndrome described by Amarenco in 1987 (1), and awareness of it is essential for surgeons.

The pudendal nerve presents three terminal branches: the inferior rectal nerve, perineal nerve and dorsal nerve of the penis/clitoris (2-4) with critical entrapment points: the sacrospinous and sacrotuberous ligament, the falciform and Alcock's canal. In the etiology of entrapment, there appear professional/sporting activities that involve a seated position (5,6), perineal injuries (7), gynecological causes (8-10), pelvic radiotherapy, etc. (11-13).

The fundamental symptom is pain in the region of the nerve's distribution, uni or bilateral, which worsens throughout the day and when seated, and improves by lying down. It may be associated with proctological symptoms (constipation, feeling of rectal foreign body), urinary or sexual (14-16). Upon exploration, we highlight the Tinel sign (pain caused by compressing the nerve against the ischial spine) and the Rolling Test (displacement of the skin and subcutaneous layer of the anus towards the pubis, producing pain) (17).

St. Mark's distal motor latency test is of significant importance (18-22). There exist diagnostic criteria such as Nantes criteria (23) (Table I). Differential diagnosis is established with endometriosis, tumors, metastasis, etc. (23,24).

Treatment includes symptomatic pain with drugs (25-27), corticosteroid/anesthetic infiltrations (28-30), pelvic physiotherapy (31), botulinum toxin (32), etc. In surgical treatment, we highlight the transgluteal, transperineal or transvaginal technique (33-36) with the goal of freeing the nerve.

Research is being carried out into stem cells, pulsed radiotherapy, laparoscopy, etc., with as yet inconclusive results (37).

MATERIAL AND METHODS

We present a prospective observational study of 53 patients with proctalgia who meet Nantes criteria (33),

diagnosed with PNE, in whom the nerve was punctured according to technique described below.

Goals:

1. Establish the relationship between proctalgia and PNE.
2. Check the immediate response to anesthetic infiltration.
3. Evaluate post-infiltration clinical results and the treatment's effectiveness in the medium term (6 months).

To evaluate the resultados we use the Visual Analog Scale of pain (VAS) at three points:

- Baseline pain level.
- Immediately post-puncture.
- Evaluation as from 6 months.

The technique used is puncture/hydrodistention with corticosteroids-ropivacaine at 0.2% at the level of the trunk and terminal nerve branches. In lithotomy position, transdermal via, guided by transanal ultrasound scan and using a neuroestimulator. The puncture was repeated after one week from the first and after two weeks from the second puncture up to a total of 3 infiltrations (Figure 1). In patients without significant improvement, a second "round" of punctures was considered.

We have considered two working groups, whose cut-off point point was established at random according to our results:

- **Non-responders:** patients whose reduction on the VAS score as from 6 months from the first puncture was less than 30%.
- **Responders:** patients whose reduction in VAS score as from 6 months from the first puncture was greater than or equal to 30%.

Statistical analysis

The Chi-square, Cramer's V and Wilcoxon tests have been applied to the variables regarding sex, personal background, affected branches, clinical signs, positivity or negativity in the St. Mark's test and VAS score pre and post-infiltration as regards being a responder or non-responder.

TABLE I
DIAGNOSTIC NANTES CRITERIA FOR PNE

<i>Nantes Criteria</i>	1) Pain in the anatomical territory of the pudendal nerve
	2) Worsened by sitting
	3) The patient is not woken at night by the pain
	4) No objective sensory loss on clinical examination.
	5) Positive anesthetic pudendal nerve block



Fig. 1. Image of the hydrodistention puncture used in the study.

RESULTS

Out of the 53 patients studied, 43.39% are male and 56.61% female. 34.78% of the males were responders and 43.33% of the women. As regards the nerve branches affected (as they present symptoms of anterior sexual branch, middle urological branch, posterior proctological branch), 64.15% had 3 branches affected, 30.18% 2 branches affected and a single effect to the posterior branch in 5.67% of cases. We found that in the group with 3 branches affected, there was a larger number of non-responders with regard to the number with 2 branches. The median age of the studied patients was 56.8 years old (24-85 years old). As regards personal background, in the male group, we highlight antecedents of anorectal surgery (21.74%) and without interest (34.78%), and in the case of women, these two major groups included gynecological antecedents (33.33%) (Figures 2 and 3). The Tinel sign was positive in 98.1% of cases, and the Rolling Test was only affirmative in 37.73% of cases, without differences between responders and non-responders. The distal motor latency test of the nerve was pathological in 88.46% of cases and we found that in the responder group there were almost two times the number of patients (19.05%) with a normal test.

The median **pre-puncture** score on the VAS was 7.74 (median of 7.08 in the responder group and 7.83 among non-responders). The VAS score referred immediately

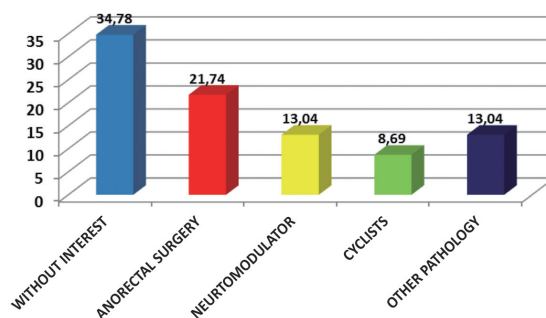


Fig. 2. Personal background of the male group.

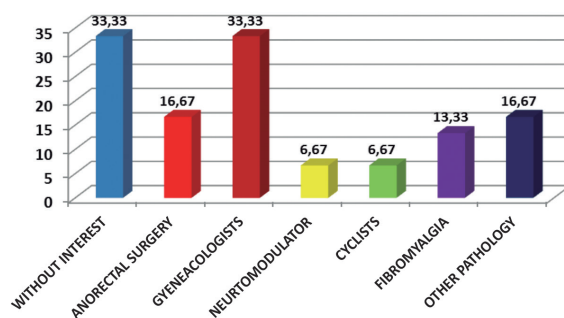


Fig. 3. Personal background of the female group.

after puncture scored a median of 1.19, finding 79.25% of patients scoring 0 and a median of 0.4 was found in responders and 1.72 among non-responders.

As from 6 months, we observed 32 non-responding (60.38%) and 21 responding (39.62%) patients (Figure 4). 24.52% of patients reported VAS 0. The median VAS score in this period was 5.39, observing in the responding group a median of 1.14 and in the non-responders 8.18 (Figure 5).

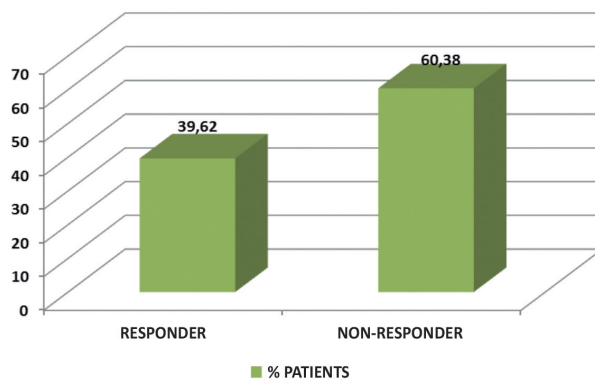


Fig. 4. Distribution of patients in responders and non-responders.

In the 159 punctures carried out, we only found 4 complications (2.51%): 2 amenorrhoeas, 1 case of vaginal bleeding and 1 vasovagal syncope. At present, 16 patients are receiving a second “round” of infiltrations.

Applying the statistics to the sex variable, background, branches affected, clinical signs, St. Mark’s in relation to the response to the treatment, we have not seen statistical significance. Conversely, upon relating the pre-infiltration VAS score with the post-infiltration score as from 6 months, both the overall sample and distinguishing between sexes, we have observed statistical significance using the Wilcoxon test (p 0.00003).

DISCUSSION

PNE is a syndrome described for the first time in 1987 by Dr. Amarenco, giving it the name Cyclists’ Perineal Paralysis Syndrome(1). Since then, Shafik, Baurtant, Robert, Beco, Benson, etc., have cast light on different aspects related with the syndrome, despite the fact that there are many questions to clarify (2).

The published series assert that incidence and prevalence are unknown, probably because this pathology is underdiagnosed, with the median time of diagnosis standing at 4 years (range from 1 to 15 years). During this time, patients are evaluated by several specialists (surgeons, gynecologists, urologists, etc.) who often, due to ignorance of this syndrome, do not issue proper diagnosis, offering ineffective solutions, so the time of diagnosis is prolonged, worsening the patients’ quality life and increasing the unnecessary cost of improperly managed healthcare resources.

As regards distribution by sexes, Motto (38) states that PNE appears more frequently in the female sex, stating that in coming years in Europe and America, 1 of every 7 women will develop PNE, probably because a large part of the etiological factors are characteristic of the female sex, in line with the long list of gynecological causes (39). The results of our

study coincide with what has been published, as 56.6% of our patients are women and we found that in the group of males 34.78% responded to the infiltrations, while in the group of women, 43.33% responded, so we deduce that women respond better to anesthetic infiltrations although without statistical significance.

The published series assert that in PNE there is no pre-dilection for any age group, an aspect we confirm in our sample (40).

Etiology suggested professional or sporting activities that involved long periods in a seated position, such as horse-riding, cycling, etc. (1,6). In our sample, there are 4 professional cyclists, who represent 7.5% of cases. There are gynecological-obstetric causes, such as vaginal childbirth, episiotomy, pelvic and vaginal surgery (8-10) where a greater incidence of PNE has been observed. In our series, we found 33.3% of women with gynecological antecedents. In relation with anorectal surgery, few studies analyze its relationship with PNE, although Baurtant (41) asserts that pelvic surgery is one of the most frequent causes in the syndrome. In our series, 18.86% of patients had antecedents of anorectal surgery.

Other processes related with the syndrome are perineal injuries (7), pelvic radiotherapy, rectal prolapse, etc., which we have not objectively represented in our sample.

There are many cases where no prominent cause is identified. Accordingly, in our sample, 33.33% of women and 34.78% of men did not present antecedents of interest.

In our study, proctalgia is the fundamental constant to which symptomatology of other nervous branches may be added (14-17). Beco, in 2004 (18), analyzed the association between fecal and urinary incontinence and perineal pain, observing that 71% of patients presented two of these symptoms while 13% presented all three. Conversely, in our sample, 64.15% of cases presents symptomatology of 3 branches, 30.18% affecting 2 branches and 5.67% only affecting the posterior branch. We found that in the group of patients with symptomatology of the three branches there was a larger percentage of non-responder patients (65.71%) compared with those who were affected in 2 branches (50%), so we observed a poorer response among patients affected in 3 branches.

The Tinel sign is considered a valuable, very sensitive sign, since in 98.1% of our patients it was positive, with only patient, a non-responder, who showed this sign negative, which also demonstrates it to be a specific sign. As for the Rolling Test (17), it was positive in only 37.73% of our sample, not observing significant differences between responders and non-responders, so we consider this sign as not very sensitive and not very specific.

The most important neurophysiological study is the Distal Motor Latency Test of the nerve (St. Mark’s) which is based on measuring the speed of the nerve’s motor conduction, while not useful to evaluate the extension of the injury

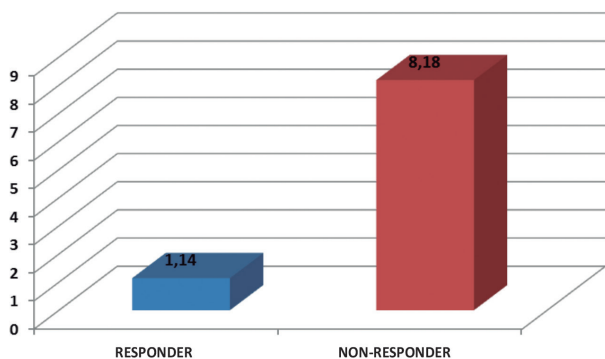


Fig. 5. Median VAS score in the group of responders and non-responders as from 6 months.

(19,20). In our sample, in 88.46% of cases it was pathological. If we distinguish between responders and non-responders, among the first it was pathological in 80.95% of cases, while among the second it revealed pathological values in 90.625%, confirming that it is a sensitive, specific test. In view of our data, we deduce that when patients present a normal St. Mark's test, they respond better.

It is fundamental for a diagnosis of exclusion to carry out image tests. In our sample CT was carried out in 14 cases, MRI in all and endorectal ultrasound scan in 49 patients.

It is also necessary to make a differential diagnosis with the syndrome of entrapment of the posterior cutaneous leg nerve, as in the latter pain may affect the perineum but only the scrotum or labia majora without implying, contrary to the neuralgia of the pudendal nerve, anus, penis or clitoris (42). M. Mollo (21) applied doppler color to the internal pudendal artery to back up the diagnosis.

Treatment of PNE is based on 3 pillars, symptomatic treatment, anesthetic corticosteroid punctures and surgical intervention.

In our series, all the patients have received medical treatment, with the aid of amitriptyline and antiepileptics, such as pregabalin or gabapentin, widely described in the literature (26,27).

The basis of our study is to analyze treatment with corticosteroid-anesthetic punctures (28-30,40,43). It is a safe technique, as we have only observed 4 minor complications. With regard to surgical treatment, in Spain it is not developed, although the Hospital de Vigo is beginning with the technique of nerve decompression by the translumbar via.

We have used the VAS scale applied to proctalgia because we consider it is an easy method and that it faithfully reflects reality. We have thus obtained a median pre-puncture score of 7.74. Among responders, the median score on the VAS scale was 7.08 and among non-responders 7.83, so in the pre-puncture period we already observed a slight tendency to present a higher score among the non-responder group.

The median VAS score in the period immediately after puncture was 1.19 and it should be noted that in 42 of the 53 studied patients, the score referred was 0, from which we may deduce that the technique is effective in the short term. However, when analyzing its evolution, we observed that many patients among those who showed a favorable response, over time became symptomatic and reach a score equal to or above the pre-puncture score, so the VAS score immediately after puncture is not a good long term response indicator.

As from 6 months, after the first puncture, a median score was obtained on the VAS scale of 5.39. In 13 patients the score was 0, which represents 24.52% of cases. We obtained 32 non-responder patients (60.38%) and 21 res-

ponder patients (39.62%). From these figures, we deduce that, although the response is not complete in the long term, responders maintain considerable improvement over time after the first puncture. This fact is highly significant as many of our patients spend years with disabling symptomatology, resistant to any kind of conventional treatment, involving a range of specialists who are unable to offer a solution to their problems, so an improvement in their pain level of more than 30% in their pre-puncture VAS score represents so much relief, satisfaction and gratitude that we consider it a major advance and a solution, which although often not total, greatly improves quality of life.

Applying the Wilcoxon test and comparing values in the pre-puncture VAS score with regard to the post-puncture VAS score as from 6 months, we observed that VAS values dropped significantly, which confirms that the corticosteroid-anesthetic hydrodistention puncture is an effective technique that offers full improvement and in other cases partially, to a significant percentage of patients.

The literature barely registers series that evaluate monitoring beyond the period immediately following the first puncture, such as ours. Accordingly, Ricci (40) analyzes infiltration of the pudendal nerve guided by CT in a series of five patients, asserting that all reported reduced pain after infiltration, but adding that only monitoring would provide evidence of maintenance of good results. Similar findings were obtained by McDonald and Spigos (44) without obtaining long term data. Fannucci (45) coincides with us in that punctures are a safe treatment, obtaining greater clinical effectiveness than us (92% at 12 months). Dehkharghani (46) showed a high rate of success, but based on the result immediately following infiltration without providing long-term data. In 2009, Filler (47) advocates the use of MRI to obtain precise images of the nerve and to better manage infiltrations. As we mentioned above, our technique is guided by ultrasound scan. Rofael in 2008 (34) showed that ultrasound scan allows visualization of anatomical points, and observation of the local anesthetic's diffusion, improving the technique's precision, reducing complications to a minimum. Choi (48) applied fluoroscopy to improve vision, returning a success rate of 100% based on the reduction of 2 points in the VAS scale, measured at 12 months after puncture. In our opinion, we should point out that a reduction of 2 points in the VAS scale has not been considered as successful, as our proposition is a percentage reduction in the baseline score in the VAS scale, considering this a better reflection of reality. Le Tallec de Certaines (49) analyzed the time of pain evolution and response to the infiltration, observing better results in the cases of neuralgia with a start of lesser pain at 1 year. Analyzing this fact, it may be due to the fact that short-evolution "acute" pain, is caused by solvable, transitory reasons, while chronic pain may be due to permanent causes with difficult resolution.

At present, two trends exist after failure of the first puncture. These are, new “round” of punctures or referral to decompressive surgery.

In our case, we opted for a second “round” of punctures, as this is less aggressive than insisting on a conservative treatment.

Popeney, Ansell and Renney (50) advocate decompressive surgery, reporting positive results shown by fifty patients. In 2007, Robert (51) provided better results with surgical treatment vs. medical treatment.

In some specialized pain units, the use of pulsed radio-frequency or the use of sacral neuromodulators are being researched, which open new lines of investigation.

In conclusion, PNE is a painful, disabling syndrome, whose incidence and prevalence are unknown, under-diagnosed and more frequent among the female sex with no age range for its appearance. There exists a high percentage of patients with gynecological antecedents, anorectal surgery and without interest. According to our data, the affectation of three branches is the most frequent and the group that responds worst. The Tinel sign is sensitive and specific, unlike the Rolling Test and we consider that the St. Mark's test is a good diagnostic test. In our experience, treatment with corticosticoid-anesthetic hydrodistention puncture manages to rescue/improve a major percentage of patients, with good results in the medium term (6 months) relieving pain in 39.62% of cases and in a statistically significant manner as regards pre-puncture pain.

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CONFLICTS OF INTEREST

The authors state they have no conflicts of interest.

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