



Diagnostic Criteria for Pudendal Neuralgia by Pudendal Nerve Entrapment (Nantes Criteria)

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Aims: The diagnosis of pudendal neuralgia by pudendal nerve entrapment syndrome is essentially clinical. There are no pathognomonic criteria, but various clinical features can be suggestive of the diagnosis. We defined criteria that can help to the diagnosis. **Materials and Methods:** A working party has validated a set of simple diagnostic criteria (Nantes criteria). **Results:** The five essentials diagnostic criteria are: (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. Other clinical criteria can provide additional arguments in favor of the diagnosis of pudendal neuralgia. Exclusion criteria are also proposed: purely coccygeal, gluteal, or hypogastric pain, exclusively paroxysmal pain, exclusive pruritus, presence of imaging abnormalities able to explain the symptoms. **Conclusion:** The diagnosis of pudendal neuralgia by pudendal nerve entrapment syndrome is essentially clinical. There are no specific clinical signs or complementary test results of this disease. However, a combination of criteria can be suggestive of the diagnosis. *NeuroUrol. Urodynam.*
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INTRODUCTION

After having been completely neglected and poorly defined for a long time, the diagnosis of pudendal neuralgia^{1,2} is now fairly easy to establish in the presence of typical perineal pain, previously considered to be psychogenic due to the absence of organic lesions demonstrated on imaging or endoscopy.³ The price of this success is that this entity is increasingly over-diagnosed or simply diagnosed by default in the presence of pelvic, perineal and buttock pain and in the absence of a diagnosis of organic disease. All forms of pain accentuated by sitting tend to be attributed to pudendal neuralgia. Although clinical neurophysiology has considerably improved our knowledge of this disease, its limitations have also been defined; it must therefore be considered to be a complementary investigation, but can never be used to formally confirm or exclude the diagnosis of pudendal neuralgia.⁴

We have therefore tried to define diagnostic criteria for pudendal neuralgia by pudendal nerve entrapment that do not pretend to cover all clinical situations, especially as the expression of pain is eminently variable and this type of pain is particularly complex as it is often associated with multiple, perplexing functional symptoms. The objective of this study was to elaborate and publish a limited number of simple criteria designed to avoid excessive or incorrect diagnosis of pudendal neuralgia. All doctors concerned with the perineum should be familiar with these criteria, but, due to their inevitably oversimplistic nature, they may need to be reconsidered on a case by case basis by experts depending on the clinical context.

These criteria were discussed and validated by a multi-disciplinary working party in Nantes (France) on 23 and 24 September 2006 (Nantes criteria) and then by members of

the Club d'électrophysiologie périnéale (Francophone perineal electrophysiology club). These proposed criteria were approved by the SIFUP PP (*Société Interdisciplinaire Francophone d'Urodynamique et de Pelvi-Périnéologie*).

In the absence of pathognomonic imaging, laboratory and electrophysiology criteria, the diagnosis of pudendal neuralgia, like any form of neuralgia, remains primarily clinical and empirical and must constantly be reviewed in the light of the clinical course. Pudendal nerve entrapment (PNE) is the most frequent aetiology and is also established on the basis of elements of clinical suspicion. Other aetiologies have also been described:⁵ post-herpetic neuropathy, stretch neuropathy (although it is usually not painful or only very slightly painful), peripheral polyneuropathy, post-radiotherapy neuropathy, neoplastic compression, etc. In fact, only the operative finding of nerve entrapment and post-operative pain relief⁶ can formally confirm the diagnosis of pudendal neuralgia due to nerve entrapment (except for a possible placebo effect of surgery).

Four diagnostic domains have been defined:

- essential criteria for the diagnosis of pudendal neuralgia
- complementary diagnostic criteria
- exclusion criteria
- associated signs not excluding the diagnosis.

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ESSENTIAL CRITERIA FOR THE DIAGNOSIS OF PUDENDAL NEURALGIA BY PNE

These five criteria are considered to be essential and must therefore all be present in order to conclude on a diagnosis of pudendal nerve entrapment syndrome or compressive pudendal neuralgia.

Pain in the Territory of the Pudendal Nerve: From the Anus to the Penis or Clitoris

This nerve trunk pain must be situated in the territory of the pudendal nerve, which extends from the anus to the clitoris or penis (Fig. 1). Pain may be superficial or may be situated slightly deeper in the anorectal region, vulvovaginal region and distal urethra. This criterion excludes pain exclusively confined to the coccygeal region, sacrum, buttocks, pubis and hypogastric region, but the pain of pudendal neuralgia can be referred to these zones. Although scrotal skin is innervated by sacral nerve roots and the pudendal nerve, the testis (and ovary), epididymis and vas deferens are innervated by thoracolumbar nerve roots.

Pain is Predominantly Experienced While Sitting

This is an essential clinical feature providing evidence in favor of the hypothesis of nerve compression in the context of an entrapment syndrome. Nerves must be freely mobile to avoid compression during movements (example of the ulnar nerve at the elbow) or during the pressure of sitting in the case of the pudendal nerve. Any loss of mobility of the nerve (regardless of its site) is therefore associated with a risk of compression against rigid ligamentous structures such as the falciform process of the sacrotuberous ligament.⁷ Pain is due to excessive pressure and not to the sitting position, as clearly illustrated by relief of pain when sitting on a toilet seat (provided the patient sits for a sufficiently long time). This dynamic aspect is essential, as if the pain were exclusively related to a compression phenomenon, it would be continuous (although pain secondary to a tumor and experienced while standing or in bed can also be aggravated by sitting). Very often, pain is initially experienced only in the sitting position, but with time pain tends to become much more continuous even if it is still predominantly experienced while sitting.

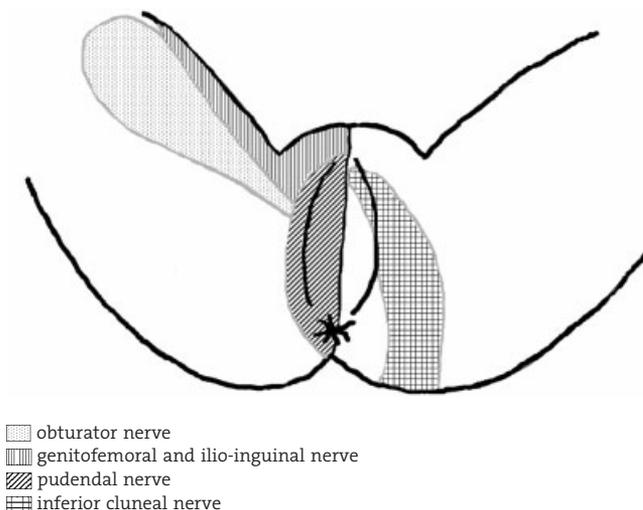


Fig. 1. The innervation of the perineum.

The Pain Does Not Wake the Patient at Night

This criterion is the direct consequence of the previous criterion. Many patients experience pain at bedtime and may have difficulties going to sleep. Although they can be woken by associated symptoms (e.g., need to urinate), they are never woken by perineal pain. Patients only exceptionally report a history of waking due to pain at night, but these episodes are only transient.

Pain With No Objective Sensory Impairment

This is an essential clinical finding. The presence of a superficial perineal sensory deficit is highly suggestive of a sacral nerve root lesion, particularly involving the cauda equina nerve roots, or a sacral plexus lesion. These proximal lesions usually do not cause pain and present clinically with sensorimotor deficits, especially sensory loss and sphincter motor disorders. Several hypotheses can be proposed to explain this absence of objective sensory impairment. The compression may be insufficient to induce a lesion of the fibers of superficial sensation, as observed in the case of sciatica and many cases of carpal tunnel syndrome. It may also have an anatomical explanation, as several anatomical territories overlap at this level: the territory of the pudendal nerve, the territory of the posterior femoral cutaneous nerve and its inferior cluneal branches, and the territories of nerves arising from the first lumbar nerves (especially ilioinguinal and genitofemoral).⁸

Pain Relieved by Diagnostic Pudendal Nerve Block

Anesthetic infiltration of the pudendal nerve⁹ significantly relieves pain for the duration of local anesthesia. This is an essential criterion, but is not specific as it simply indicates that the pain is situated in the territory of the pudendal nerve; pain related to any perineal disease (e.g., anal) would also be relieved by pudendal nerve block and other types of nerve lesions would also have a positive diagnostic block when they are situated distal to the site of infiltration. A negative block does not formally exclude the diagnosis when it is not performed with sufficient precision or when it is performed too distally (e.g., in the pudendal canal, while the pudendal nerve lesion may be situated at the ischial spine). The infiltration technique (with or without CT guidance, with or without neurostimulation) has only a minor impact on the positive or negative response to nerve block.

COMPLEMENTARY DIAGNOSTIC CRITERIA

Burning, Shooting, Stabbing Pain, Numbness

Pudendal neuralgia presents the characteristics of neuropathic pain, which is described as burning, shooting, stabbing or aching pain and numbness, although more than four criteria of the DN4 are only rarely present¹⁰ (clinician-administered questionnaire named DN4 consisting of both sensory descriptors and signs related to bedside sensory examination, comprises 10 criteria, 4 of which must be present for the diagnosis of neuropathic pain).

Allodynia or Hyperpathia

Allodynia or hyperpathia, highly suggestive of neuropathic pain, correspond, in the pudendal nerve territory, to intolerance of tight clothes and underwear (boxer shorts are preferred to briefs), and intolerance of vulval contact (as in vestibulodynia) with superficial dyspareunia.

Rectal or Vaginal Foreign Body Sensation (Sympathalgia)

Patients generally use fairly vivid terms to describe their deep pain, generally situated in the anus and rectum or sometimes in the vagina or urethra. They frequently describe a feeling of “foreign body,” but other expressions are also suggestive: feeling of a stake, a lump, heaviness, a tennis ball, a gnawing, or crawling feeling. This symptom is sometimes incorrectly called levator ani syndrome in the absence of any correlation with levator ani hypertonia.¹¹ This pain has an autonomic connotation and temporary relief of these sensations by anesthetic block of sympathetic fibers of the ganglion impar suggests that they are mediated by sympathetic fibers, justifying the term “sympathalgia.”

Worsening of Pain During the Day

Absence of pain in the morning on waking, slight pain in the morning, deterioration during the day reaching a peak in the evening until the patient goes to sleep is a very characteristic temporal profile of pudendal neuralgia.

Predominantly Unilateral Pain

Perineal pain is particularly suggestive of a pudendal nerve trunk lesion when it is unilateral (and when it is experienced in all of the anterior and posterior hemiperineum), but midline or central pain does not exclude the diagnosis.

Pain Triggered by Defecation

This is a feature of predominantly posterior pain; pain is not experienced immediately after defecation, but generally several minutes to one hour later.

Presence of Exquisite Tenderness on Palpation of the Ischial Spine

Palpation of the ischial spine (posterior and slightly lateral) during digital rectal or vaginal examination is very often tender. This does not constitute a true Tinel sign, as this palpation is locally tender, but does not trigger the distal pain described by the patient. Many anatomical structures are situated at this level, making this tenderness very difficult to interpret: passage of the pudendal nerve in the sacrospinous ligament, insertions of the sacrospinous ligament, ischiococcygeal fibers of levator ani muscles, diffuse hypersensitivity. Furthermore, tenderness of this region is not specific as it is also observed in asymptomatic subjects. However, unilateral tenderness at this site is suggestive.

Clinical Neurophysiology Findings in Men or Nulliparous Women

Childbirth is the commonest cause of stretch injury to the pudendal nerve, eliminating the specificity of this examination when it is performed in multiparous women. However, clinical neurophysiology data can be useful in men and nulliparous women in the absence of a history of constipation, surgery, or known proximal spinal cord or nerve root lesions.

EXCLUSION CRITERIA

Exclusively Coccygeal, Gluteal, Pubic, or Hypogastric Pain

This type of pain does not correspond to the anatomical territory of the pudendal nerve.

Pruritus

Pruritus is primarily suggestive of a dermatological lesion (atrophic lichen planus, etc.) rather than a nerve lesion. The DN4 criteria for neuropathic pain comprise “prickling,” as this term may be used by patients, but the concept of pruritus includes a need to scratch which is not experienced in the context of pudendal neuralgia.

Exclusively Paroxysmal Pain

Paroxysmal shooting pain with neuropathic features is suggestive of a compressive lesion, but justifies further investigation by imaging of the pelvic region (pudendal nerve neurofibroma or schwannoma), cauda equina (sacral schwannoma) and spinal cord (meningioma). *Proctalgia fugax* is sufficiently suggestive not be confused with pudendal neuralgia, but is not sufficiently well known. *Proctalgia fugax* is predominantly anorectal, essentially nocturnal, recurrent pain that can last several minutes to one hour, with attacks that can occur several times a year for many years, without deterioration. Although some authors have proposed a neurological aetiology, the great majority of cases are strictly idiopathic with a controversial pathophysiology (smooth muscle spasm?).^{12,13} This type of paroxysmal pain also comprises pain experienced exclusively during defecation (suggesting a proctological disease), or micturition (suggesting a urological disease), or related to sexual intercourse (vaginismus, vestibulitis).

Imaging Abnormalities Able to Account for the Pain

Medical imaging is not contributive to the positive diagnosis of pudendal neuralgia, but can be useful to exclude other diagnoses. However, imaging may reveal an intercurrent disease clearly unrelated to the neuralgia, for which treatment will not modify the course of the neuralgia. The finding of arachnoid cysts remains a difficult problem, but these cysts are classically considered to be asymptomatic. In any case, they cannot be considered to be responsible for pudendal neuralgia. Imaging is therefore essential, whenever the clinical features do not strictly meet the diagnostic criteria described in this article and will be decisive when it demonstrates a lesion able to account for the pain (especially a nerve tumor).

ASSOCIATED SIGNS NOT EXCLUDING THE DIAGNOSIS

The symptoms of pudendal neuralgia may be strictly limited to the diagnostic criteria defined above, but many patients present associated, polymorphic and perplexing symptoms that are often difficult to attribute to the pudendal nerve. Clinical experience and the course of these symptoms in response to treatment indicate that these signs do not exclude the diagnosis, although they may be difficult to explain.

Buttock Pain on Sitting

Gluteal innervation is not dependent on the pudendal nerve and isolated buttock pain, even occurring while sitting, cannot be considered to be due to pudendal neuralgia. However, the combination of perineal neuralgia and buttock pain can be explained by a common, fairly proximal conflict underneath the piriformis muscle with a concomitant lesion of the posterior femoral cutaneous nerve or inferior gluteal nerve. Buttock pain can be related to trigger points or spasm of deep gluteal muscles: obturator internus

and piriformis muscles, possibly due to reflex muscle contractures secondary to pain or regional myofascial syndrome, extremely common in this context (reflecting regional hypersensitivity).

Referred Sciatic Pain

More or less truncated sciatica is extremely frequent in the context of pudendal neuralgia and can be explained by a lesion of the posterior femoral cutaneous nerve or sciatic trunk (with or without piriformis or obturator internus syndrome).^{14,15} Central hypersensitivity phenomena can also be involved in view of the convergence between S1, S2, and S3 spinal levels (see effects of posterior tibial nerve stimulation on overactive bladder or perineal pain).

Pain Referred to the Medial Aspect of the Thigh

Pain referred to the obturator nerve territory may reflect an obturator internus syndrome, as this muscle is in contact with the nerve.

Suprapubic Pain

Suprapubic pain can be due to hypertonia of the puborectalis component of the levator ani muscles. Bone tenderness may suggest a complex secondary pelvic pain syndrome (low-grade reflex sympathetic dystrophy).

Urinary Frequency and/or Pain on a Full Bladder

Urinary frequency is often associated with pudendal neuralgia and tends to evolve intermittently, in parallel with the pain, allowing it to be attributed to pudendal neuralgia rather than to bladder dysfunction. There are probably synaptic interconnections associated with inappropriate processing of nociceptive messages resulting in transmission of false urges. Patients sometimes report urethral or hypogastric pain worsened by bladder filling and relieved by voiding. Such symptoms should be documented by a voiding diary; when voided volumes are small and very constant, cystoscopy under general anesthesia should be performed with bladder distension, looking for signs of painful bladder syndrome/interstitial cystitis. In contrast, very variable voided volumes are suggestive of detrusor hyperactivity, possibly part of central hypersensitivity and reflex autonomic phenomena.

Pain Occurring After Ejaculation

This isolated symptom, which is perplexing in the absence of infection (absence of seminal vesiculitis), cannot be attributed to pudendal nerve entrapment syndrome. However, it is fairly frequently associated with pudendal neuralgia and can be considered to reflect regional hypersensitivity.

Dyspareunia and/or Pain After Sexual Intercourse

The frequency of sexual intercourse is often reduced in the context of pudendal neuralgia, essentially because chronic pain decreases libido. Sexual intercourse is rarely very painful, but pain can be experienced in the case of vulval allodynia, but patients generally do not report pain during intercourse, but worsening of pain over the hours following intercourse.

Erectile Dysfunction

Pudendal neuralgia, affecting a somatic nerve, is only partially involved in erection. Classically, the main sexual function of the pudendal nerve is sensory (dorsal nerve of the penis and dorsal nerve of the clitoris), but it is also involved in pre-ejaculatory hyperrigidity and in the clonic nature of ejaculation. Patients with pudendal neuralgia frequently complain of a feeling of penile numbness, decreased sexual sensations or even decreased penile rigidity. Analgesic drugs can also have a negative impact on erection and ejaculation.

Normal Clinical Neurophysiology

Electrophysiological studies (electromyography and nerve conduction studies) only investigate large motor fibers and may not detect selective lesions of small sensory fibers. Furthermore, due to the postural nature of the pain, a neurological lesion may not always be present in the context of intermittent conflict.

CONCLUSIONS

The diagnosis of pudendal neuralgia by pudendal nerve entrapment syndrome is essentially clinical. There are no pathognomonic criteria, but various clinical features can be suggestive of the diagnosis. In the presence of the 4 essential clinical diagnostic criteria (pain in the territory of the pudendal nerve, worsened by sitting, the patient is not woken at night by the pain and no objective sensory loss) a diagnostic anesthetic pudendal nerve block should be performed and a positive block strongly supports these elements of clinical suspicion (5th criteria). However, pudendal pain is complex and interpretation of the various signs that are often

TABLE I. Diagnostic Criteria for Pudendal Neuralgia by Pudendal Nerve Entrapment

Essential criteria
Pain in the territory of the pudendal nerve: from the anus to the penis or clitoris
Pain is predominantly experienced while sitting
The pain does not wake the patient at night
Pain with no objective sensory impairment
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 on 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

associated may improve the understanding and management of this disease (Table I).

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