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Pudendal nerve neuralgia: Awakening to an underestimated reality

Marco Orsini, Jacqueline Fernandes Nascimento, Adalgiza Mafra Moreno, Antônio Marcos da Silva Catharino, Luciana Armada

ABSTRACT

Introduction: Pudendal neuralgia is the constituent pain component of the pudendal syndrome, caused by pudendal neuropathy. Described as a chronic and severely disabling neuropathic perineal pain, it is often underdiagnosed and inadequately intervened. The present case report intends to emphasize a rare pathology, some of its clinical aspects and diagnostic criteria, demonstrating an example of how the delay in diagnosis can lead to an excessive expenditure of health resources, as well as to significant physical and psychological suffering.

Case Report: We report a case of a 45-year-old male pianist, who reported excessive workload in a sitting position and denied personal history of comorbidities. He reported that about eight months ago he started having unilateral, strong pain in the penis. He stated that such pain presents as tingling, pricking, throbbing, and sometimes burning. It typically worsened when he sat down, relieved with walking, and gradually increased during the day and worsened at night.

Conclusion: Importantly, patients must be encouraged to prevent painful stimuli and to actively participate in physical therapy, since chronic pain represents a mental and economic burden for the patient. Lifestyle changes are one of the primary elements of the therapeutic plan, and although retrospectively this diagnosis seems simple, it is underestimated and most patients are mistreated.

Keywords: Neuralgia, Neuropathic pain, Pelvic pain, Pudendal nerve

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INTRODUCTION

Pudendal neuralgia is the painful component of the pudendal syndrome, caused by pudendal neuropathy. This condition affects both sexes in a similar way, and in children it is the result of congenital abnormalities of the nerve pathway. It is described as a chronic and severely disabling neuropathic perineal pain, which is usually underdiagnosed and inadequately intervened [1]. It is often associated with significant losses in quality of life, in terms of its subjective and multidimensional aspects: cognitive, behavioral, sexual, emotional, and psychosocial [2].

The pudendal nerve is a mixed nerve with sensory, motor, and autonomic functions that originates from the sacral plexus and is formed from the spinal nerve roots S2–S4. It runs anterior to the piriformis muscle and then passes between the sacrotuberous and sacrospinous ligaments, which are analogous to a

pincer, and which can sometimes "pinch" or impinge the nerve. Upon leaving this site, the nerve travels through the pudendal canal—Alcock's canal—and divides into the perineal nerve, the dorsal nerve of the penis or clitoris, and the inferior rectus nerve [3]. The dorsal nerve innervates the penis and the clitoris. The inferior rectal nerve innervates the external anal sphincter and the perianal skin. The perineal nerve innervates the bulbospongiosus, ischiocavernosus and levator ani muscle, and sends sensory branches to the skin of the labia majora and scrotum. Furthermore, many variations in nerve structure are reported [4].

It is important to point out that due to the little—and almost null—knowledge about this condition, the patients affected by it are submitted to exhaustive and expensive interventions. Even so, it is observed with certain frequency, the dark and Jurassic diagnosis of hysteria, as well as cauda equina syndrome, urinary tract infection, among others commonly used [5, 6].

Effectively, there are five criteria considered essential that must be present at the same time to arrive at the diagnosis of pudendal neuralgia, which are: (1) pain in the area of the pudendal nerve, from the anus to the penis or clitoris; (2) the pain is predominantly felt when the user is sitting, with the pain being due to excessive pressure on the nerve rather than the sitting position, as illustrated by the pain relief when the user sits on the toilet; (3) pain does not wake the patient at night (only exceptionally is a history of nocturnal skin awakenings reported, but these episodes are rare); (4) pain without objective decrease in sensitivity; and (5) pain is relieved by pudendal nerve block [7].

Therefore, the aim of the present study is to highlight a condition that, although rare, causes significant physical and psychological detriment. The symptoms presented by the patient may confuse and/or mimic other conditions, hinder timely diagnosis, lead to a disproportionate expenditure of health resources, and lead to the establishment of therapies that are ineffective and sometimes not directed at the problem.

CASE REPORT

A 45-year-old male pianist reported excessive workload in a sitting position and denied personal history of comorbidities. He reported that about eight months ago he started having unilateral, strong pain in the penis. He stated that such pain presents as tingling, pricking, throbbing, and sometimes burning. It typically worsened when he sat down, relieved with walking, and gradually increased during the day and worsened at night. The narrative of pain spreading to other regions was sporadic, being the localized pain his biggest and most relevant complaint. He denied triggering factor or event of the symptoms. In addition to the worsening of painful symptoms, he noticed a decrease in libido and absence of spontaneous erections.

The path of neuropathic pain started in the region from the anus to the penis in most cases; although our patient's main complaint was topographically located at the base of the glans, it also covered the superolateral surface. For maintenance of the complaints he was evaluated by several specialists who considered the problem to have a urological cause. During urination he felt uncomfortable and apprehensive due to frequent urethral burning, sometimes with the sensation that a foreign body was occupying this region. Another point that deserves to be highlighted and reinforces our hypothesis is the attenuation of pain when the patient promoted release from nerve compression, such as standing up, stretching, and especially sitting in places that do not cause compression (toilet seat). Ejaculation became difficult to manage due to the fear of a possible neuropathy trigger. Urgency of voiding was also reported. During the neurological examination allodynia had occurred in the perineum and glans homolateral to the lesion. The patient had significant improvement of pain and paresthesias after about two months of treatment with Gabapentin 300 mg—3×/day and Duloxetine 60 mg—1×/day.

The complementary exams performed were complete blood count, urine culture, and magnetic resonance imaging of the pelvis and lumbar spine, all of them without significant alterations. Due to the specificity of the complaints, we did not think it necessary to perform an electroneuromyography of the pudendal nerve. Electrophysiological studies can help in the diagnosis, among them we can mention sensory and motor evoked potentials and electroneuromyography of the rectal and urinary sphincters. We chose not to perform them due to the specificity of the clinical picture.

DISCUSSION

Pudendal neuralgia should be suspected in patients with a history of pelvic pain, particularly perineal and genital, with or without associated sexual, urinary, or bowel symptoms. The pain often presents with subtle onset, except when generated by acute trauma. It usually subsides in the morning and progresses throughout the day. In general, patients complain of burning pain, tingling pain, stabbing pain, and shock-like pain. In more than 50% of patients, the pain is exacerbated by sitting and relieved by standing, lying down, or sitting on a toilet seat [8].

Several are the symptoms presented by patients, among them there are intestinal, urinary and sexual manifestations with or without pain. The distribution of pain may be limited or extensive and may include vulva, vagina, clitoris, perineum, and rectum in women; glans penis, scrotum, perineum, and rectum in men. In addition, coccygeal pain, referred pain in the calf, foot, and toes have been described in the literature. Allodynia is also found as a frequent complaint, often described as

pain or discomfort on contact with garments. This is an indicator of central sensitization. Pain can also be felt outside the pudendal nerve innervation territory and may include vague, neuropathic pain in the intra-abdominal region, posterior, and inner thigh or lower back [7, 9].

Other symptoms associated with pudendal neuralgia include urinary frequency, urgency, symptoms mimicking interstitial cystitis, painful nocturnal orgasms and ejaculation, dyspareunia, persistent sexual arousal, foreign body sensation or weight in the rectum or vagina, pain that is more prominent posteriorly and unilateral and triggered minutes or hours after defecation, and abnormal results on neurophysiological tests [10].

Pudendal neuralgia can arise as a result of mechanical or non-mechanical injury. Mechanical injury can be caused by compression, transaction, or stretching. Among mechanical causes, compression caused by incarceration of the pudendal nerve is the most common cause. Non-mechanical causes of pudendal neuropathy include viral infections such as herpes zoster and human immunodeficiency virus (HIV); multiple sclerosis; diabetes mellitus, among others [11].

The physical examination of these patients is relatively normal, except for the reproduction of pain. Symptoms depend on the site of compression and the diagnosis is essentially clinical, corroborated by neurophysiological tests [12, 13].

In men, the most common differential diagnosis of pudendal neuralgia is prostatitis. However, it is necessary to rule out inflammation of the prostate by evaluating prostate secretions or seminal fluid for leukocytes [14]. In women, morphological diseases of the uterus or ovaries are taken into consideration, as well as endometriosis, even without laparoscopic confirmation [15].

The intervention strategies for this condition include prophylactic behavioral changes (mitigation of pain triggering factors); pelvic floor physiotherapy; pharmacological therapy with Gabapentin, Pregabalin and tricyclic antidepressants; pudendal nerve blocks; surgical decompression—considered the best treatment for pudendal neuralgia—and neuromodulation [16].

CONCLUSION

Pudendal neuralgia is defined as pain along the pudendal nerve dermatome and can occur in both sexes. Being a pathology capable of mimicking many other organic conditions, its diagnosis usually comes after months of symptoms, as in the present case. Although numerous exams can be performed, there is a lack of knowledge if they also able to evaluate and identify this clinical entity.

Pudendal neuralgia should be suspected in patients with a history of pelvic pain, particularly perineal and genital, with or without associated sexual, urinary, or bowel symptoms. The pain often presents with subtle onset, except when generated by acute trauma. It usually subsides in the morning and progresses throughout the day. In general, patients complain of burning pain, tingling pain, stabbing pain, and shock-like pain. In more than 50% of patients, the pain is exacerbated by sitting and relieved by standing, lying down, or sitting on a toilet seat.

Importantly, patients must be encouraged to prevent painful stimuli and to actively participate in physical therapy, since chronic pain represents a mental and economic burden for the patient. Lifestyle changes are one of the primary elements of the therapeutic plan, and although retrospectively this diagnosis seems simple, it is underestimated and most patients are mistreated.

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Marco Orsini – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, 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

Jacqueline Fernandes Nascimento – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, 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

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Consent Statement

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.