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CASE REPORT

Pudendal neuralgia with rare symptoms hematuria and spermaturia

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ABSTRACT
Pudendal neuralgia is a rare entrapment syndrome. It is characterized by symptoms such as neuropathic pain, allodynia, hyperesthesia in the pudendal innervation area and erectile dysfunction. Rarely additional symptoms like hematuria and hematospermia may manifest. Currently there is no universally recognized treatment approach for pudendal neuralgia and management primarily focuses on symptom relief.
Here we present a case with rarely seen symptoms such as hematospermia and hematuria.
The patient presented with burning, stabbing and cutting pain localized in the scrotum, penis and perianal region with more pronounced symptoms experienced on the right side. Six months after the onset of pain the patient also developed symptoms of hematospermia and hematuria. Following four local lidocaine injections administered to the patient at weekly intervals, the symptoms showed a 60% improvement.
In summary diagnosing pudendal neuralgia poses a significant challenge due to its complex nature. Its treatment options remain uncertain. However this case highlights the potential effectiveness of local analgesic injections as a treatment modality.

Keywords: pudendal neuralgia, pain, spermaturia , hematuria

ÖZET
Pudendal nevralji: nadir görülen semptomlar hematüri ve spermatüri

Pudendal nevralji nadir görülen bir sıkışma sendromudur. Bu tablo pudendal sinirin sıkışması veya hasarı sonucu ortaya çıkar ve nöropatik ağrı, allodini, pudendal innervasyon alanında hiperestezi ve erektil disfonksiyon gibi semptomlarla karakterizedir. Nadiren hematüri ve hematospermi gibi ek semptomlar ortaya çıkabilir. Şu anda pudendal nevralji için evrensel olarak kabul edilen bir tedavi yaklaşımı bulunmamaktadır ve tedavi genellikle semptomların rahatlatılmasına odaklanmaktadır. Burada hematospermi ve hematüri gibi nadir görülen semptomları olan bir vaka sunuyoruz. Hasta skrotum, penis ve perianal bölgede lokalize olan yanma, saplanma ve kesici ağrı ile başvurdu. Belirtilerinin sağ tarafta daha belirgin olduğu gözlemlendi. Ağrının başlangıcından altı ay sonra hasta ayrıca hematüri ve hematospermi semptomları geliştirdi. Hastaya haftalık aralıklarla dört kez lokal lidokain enjeksiyonu uygulandıktan sonra semptomlar %60 iyileşme gösterdi. Özetle pudendal nevraljinin tanısını koymak karmaşık doğası nedeniyle önemli bir zorluk oluşturur. Tedavi seçenekleri belirsizdir. Ancak bu vaka lokal analjezik enjeksiyonların bir tedavi yöntemi olarak potansiyel etkinliğini vurgulamaktadır.

Anahtar kelimeler: pudendal nevralji, ağrı, spermatüri, hematüri

INTRODUCTION

Pudendal neuralgia (PN) refers to neuralgic pain appearing within the innervation area of the pudendal nerve. The pudendal nerve has sensory, motor and autonomic functions. It originates from sacral segments S2, S3, S4 [1]. The innervation areas include the anal region, perineum, labia and clitoris in females and the penis and scrotum in males. Symptoms manifest in these innervation areas [2-5]. Pain worsens when sitting and improves when standing or lying down [6].
Pudendal neuralgia was first described by French physical medicine and rehabilitation specialist Gerard Ameranco in 1988, who initially termed it "cyclist

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syndrome" due to its association with prolonged bicycling[7].
Its exact prevalence is unknown [6]. The incidence is found to be 1/100.000, with the most common cause being compression, stretching or trauma to the nerve. It is frequently observed in athletes and individuals who sit for extended periods [3-5]. Less commonly the other etiological factors include traffic accidents, chronic constipation, tumors or viral infections [3-5].
Diagnosis relies primarily on clinical assessment and the exclusion of alternative pathologies. Given its potential overlap with numerous pelvic region disorders diagnosis may be protracted and sometimes delayed for up to 5 years[5]. The Nantes Criteria, introduced in 2008 by Label et al., serve as a diagnostic framework for PN These criteria comprise four distinct categories: inclusion, complementary, exclusion and associated findings. To receive a diagnosis of PN all five fundamental criteria must be met while ensuring the absence of symptoms listed under the exclusion criteria[8].
Until now different treatment methods have been used .These include physical therapy, analgesic medications, pudendal nerve blockade, botulinum toxin injection, radiofrequency therapy, among others[3-5]. There are few studies regarding the use of analgesics [9].
Presented herein is a case of PN with hematuria and spermaturi an infrequent syndrome showcasing a remarkable response to localized analgesic blockade.
We observed a significant improvement in symptoms following a local analgesic blockade of the pudendal nerve.

CASE REPORT

A 52-year-old male was admitted to the our outpatient. He had burning, stabbing and cutting pain localized in the scrotum, penis and perianal region with a predilection for the right side. His symptoms started five years ago in 2018, the onset of pain followed six months of regular bicycling, initially impacting the right scrotum, the right portion of the penis and the perianal area. Subsequently similar sypmtoms but less intense manifested on the left side a few months later. The pain exacerbated during prolonged sitting, bicycling or extended driving while alleviated by standing or reclining. Due to the severity of symptoms the patient discontinued bicycling. Six months later he developed symptoms of hematospermia and hematuria. Hematuria and hematospermia aggravated after heavy lifting and bending.

The urological examination was normal.Hemogram and biochemical parameters and prostate-specific antigen (PSA) levels were normal. MRI scans of the sacroiliac and lumbosacral regions exhibited no significant anomalies except for disc bulging at L4-L5 and L5-S1 levels. Multiparametric prostate MRI revealed no discernible organic pathology correlating with the clinical manifestations. Transrectal and urinary ultrasound examinations were normal. In light of the absence of identifiable organic etiology and the escalating intensity and distribution of pain the patient was referred to the neurology clinic for further evaluation. In the neurological examination, hyperesthesia was present in the right scrotum, the right portion of the penis and the perianal area. The left side was normal. The cremaster reflex and anal reflex were normal. Following meticulous history-taking and thorough physical examination the patient was assessed according to the Nantes Criteria.
He fulfilled all five inclusion criteria, including characteristic neuralgic pain within the pudendal nerve distribution, aggravation of symptoms during sitting, undisturbed sleep despite pain, absence of sensory deficits on neurological assessment and relief upon pudendal block. Exclusion criteria were also meticulously evaluated, leading to a diagnosis of PN as per the Nantes Criteria. Before the injection, the patient's pain was assessed using the visual analog scale (VAS) and rated as 8/10.
Pudendal nerve block was executed by administering 10 milliliters (mL) of 1% lidocaine approximately 8-10 centimeters below the intersection point of a vertical line extending from the posterior superior iliac crest to the lateral edge of the ischial tuberosity and a horizontal line drawn from the lateral edge of the greater trochanter (Figure 1).
Pudendal nerve block therapy was administered in four sessions at weekly intervals. Following the blockade the patient reported a reduction in both pain intensity and distribution. Subsequent to the fourth blockade the pain was localized primarily around the ischial tuberosity with a VAS score of 4/10.

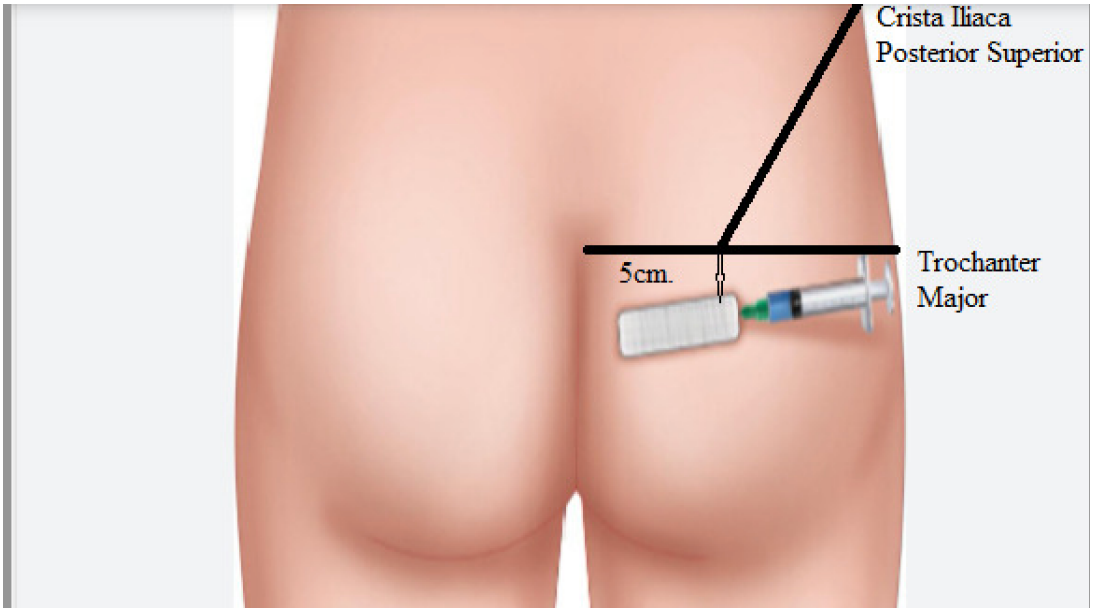

Figure 1. Application regions for pudendal nerve block

DISCUSSION

Pudendal neuralgia is frequently diagnosed with delays and this delay is attributed to inadequate history taking.

PN profoundly impacts patients' quality of life, particularly concerning sitting discomfort. While hematospermia and hematuria are rare manifestations of PN, concomitant urinary tract pathologies must be meticulously ruled out. Hematuria associated with cycling although rare warrants consideration as it may stem from physiological mechanisms like catecholamine release during strenuous physical activities such as cycling necessitating a diagnosis of exclusion.[10]

Mechanical nerve injury, viral infections or immunological factors may precipitate PN. In females common etiologies include surgical trauma, pelvic injury and childbirth, [11] while pelvic trauma predominates in males. [12]

Diagnosing PN poses challenges, primarily revolving around differentiating it from other pelvic floor-related pain pathologies. Important differentials include vulvodynia, pelvic floor tension myalgia, interstitial cystitis, and neuralgias of adjacent pelvic nerves. Comprehensive history-taking and physical examination remain pivotal in the diagnostic algorithm.

The therapeutic landscape for PN lacks standardized protocols include pelvic floor physiotherapy, analgesics, pudendal nerve blockade, botulinum toxin-A injection, surgical nerve decompression, radiofrequency and conus medullaris stimulation. [13]

Pudendal nerve blockade emerges as a promising non-invasive intervention although with limited research evidence. Notably a study by Thierry Vancaille et al. demonstrated an 86.9% reduction in pain among 66 female patients undergoing pudendal nerve blockade. [9]

In our case presentation pudendal blockade resulted in reduced pain intensity and localization in this way enhancing the patient's overall well-being. Pudendal blockade treatment was found to improve the patient's quality of life and comfort.

Pudendal neuralgia often goes undiagnosed in time especially when accompanied by atypical urological symptoms like hematospermia and hematuria. Once diagnosed using accessible interventions such as pudendal nerve blockade can significantly alleviate pain, enhance quality of life and improve patient comfort. Thus clinicians must recognize PN as a diagnosis of exclusion predicated on comprehensive clinical and examination findings.

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Ethical Considerations

The individual's participation in the study is based on voluntary consent, and informed consent has been obtained from the individual prior to the study for the use of patient information .

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