

Urethral leak: an unusual symptom of pudendal nerve entrapment

Sónia Afonso Ramos¹, Thiago Guimarães², Renaud Bollens^{3,4}

¹Hospital Prof. Doutor Fernando Fonseca, Department of Urology, Amadora, Portugal

²Centro Hospitalar Lisboa Central, Department of Urology, Lisboa, Portugal

³Centre Hospitalier Wallonie Picarde, Tournai, Belgium

⁴Centre Hospitalier Hornu / Frameries, Hornu, Belgium

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Corresponding author

Sónia Afonso Ramos

Hospital Prof. Doutor

Fernando Fonseca

Department of Urology

IC 19 2720-276 Amadora,

Portugal

phone: +351 91 672 05 26

sonia.afonso Ramos@

gmail.com

Pudendal nerve (PN) entrapment is one of the most misunderstood and underdiagnosed medical conditions. It is recognized as a cause of chronic pelvic pain syndrome. However, due to the pudendal nerve's mixed composition and complex anatomy, the presenting symptoms are varied and go beyond pain, depending on the entrapment's nature, location and duration. We report a unique case of a young patient presenting with a urethral leak refractory to antibiotics. Patient evaluation highlighted findings suspicious of pudendal nerve entrapment. The patient was submitted to a laparoscopic transperitoneal PN neurolysis, resulting in major symptoms improvement.

Key Words: pudendal nerve entrapment ◊ functional urology ◊ laparoscopic pudendal neurolysis

A 23-year-old man, with a past medical history of left testis agenesis and chronic constipation of unclear cause since childhood, presented to the urology outpatient clinic complaining of a cloudy urethral leak every day, mostly between 9 am and 11 am, for the last three months. He described defecatory strain as a worsening factor and denied a relation to sexual activity. No other concomitant symptoms such as lower urinary tract symptoms, fever, genital lesions, penile pain or pruritus were reported. Although he denied unprotected sexual intercourse, antibiotics had been previously prescribed by his general practitioner bearing in mind gonococcal urethritis, without improvement of symptoms. The patient developed a compulsive behavior of hand washing based on the obsessive belief of lack of hygiene.

Genital examination was unremarkable aside from the absence of the left testis. The urethral smear, urinalysis and urine culture showed no abnormalities. Abdominopelvic magnetic resonance imaging (MRI) was unremarkable. The suspicion of a bulbar urethral stricture was raised by a retrograde cystourethrogram. The flexible urethroscopy, on the other hand, revealed not a stenosis but a hypertonic external urethral sphincter. A digital rectal exam (DRE) was performed during the urethroscopy: the prostate was painless and normal, and no urethral discharge was triggered by prostatic massage. However, hypertonicity of the anal sphincter was highlighted, as well as tenderness of the left levator ani muscle and hyperalgesia located to the ipsilateral tendinous arch and sciatic spine. Suspecting pudendal nerve

entrapment (PNE), the patient underwent a CT-guided infiltration of the left PN with a mixture of methylprednisolone (1 ml of a 40 mg/ml solution) and bupivacaine (3 ml of a 0.25% solution). The infiltration triggered perineal pain that lasted two weeks, followed by a progressive improvement of his symptoms – the frequency of the leaks was reduced to twice a week. A diagnosis of left PNE was then supported. The patient was referred to a physiotherapist, who identified an anterior rotation of the pelvis related to a difference in leg lengths, and alterations of the vertebral alignment with increased lumbar lordosis. The patient also consulted a psychiatrist for his obsessive-compulsive behavior. Conservative management consisting of perineal hyperprotection, postural and pelvic floor physiotherapy, orthopaedic insoles and pharmacologic neuromodulation was proposed for a three month period. Despite the achieved improvement, the patient still felt quality of life impairment. A transperitoneal laparoscopic left pudendal neurolysis and fasciotomy of the Alcock canal was performed and underwent uneventfully. Four months after surgery, the patient reports no constipation, and only occasional episodes of urethral leak (less than twice a month).

DISCUSSION

More than 30 years have passed since the first literature report, by Gerard Amarenco, of symptoms suggestive of pudendal nerve entrapment in a group of cyclists [1]. Although the estimated incidence according to the International Pudendal Neuropathy Foundation is 1/100.000 [2], true numbers surely outweigh what is stated in literature, and most of the patients attempt many ineffective treatments and jump from one physician to another [3], suffering years before a correct diagnosis is done.

The origin of the pudendal nerve (PN) is in the S2–S4 sacral nerve roots and carries motor (20%), autonomic (30%) and sensory (50%) fibers [2, 4]. It divides distally in 3 branches: the inferior rectal nerve, the perineal nerve and the dorsal nerve of the penis/clitoris [2]. PNE is increasingly recognized as a cause of chronic pelvic pain [4, 5], especially after the publication of the Nantes criteria that standardized the diagnosis of pudendal neuralgia [6]. However, its mixed nature and possible anatomic variations render its lesion responsible for a wide constellation of symptoms, including overactive bladder, sexual dysfunction, and constipation [4, 5, 6]. However, this is sometimes overlooked and PNE has been neglected as a functional pathology.

This case's presentation posed a great challenge for proper diagnosis. Cystourethrogram and urethroscopy images pointed out a hypertonicity of

the external urethral sphincter, and the external anal sphincter and left levator ani muscle were also found to be hypertonic on DRE. These findings brought forth the suspicion that a compression of the PN could be causing the hypertonicity as well as bulbospongiosus muscle's spasm, as the nerve contributes to all of these muscles' innervations [4, 5, 7]. Therefore, the resulting intermittent squeezing of the Cowper's glands would lead to the leak of their secretions through the urethra.

The key aspect to support our hypothesis was the tenderness of the left levator ani muscle and pain elicited on applying digital pressure on the ischial spine and arcus tendinous. These are described in the literature as frequent signs of PNE [4, 5]. Although the exact pathophysiology is not completely understood, there is a rationale that in some cases pelvic distortion and postural misalignment (both findings in our patient's examination) lead to a reflex spasm of the pelvic floor muscles, eventually resulting in a PN mechanical compression. Pressure induced by sitting could be a cause of clonic muscle spasms and symptom triggering [2, 6], explaining our patient's 'scheduled' clinical presentation, with a peak frequency in the morning and no symptoms at night or upon awakening.

Since there are no pathognomonic electrophysiological or radiological findings, the diagnosis is mainly based on symptoms and clinical signs, and supported by the improvement of symptoms after a CT-guided infiltration of the nerve [2, 3, 5], as seen in this case. The initial treatment is always conservative with a combination of pharmacologic neuromodulation, muscle relaxants, perineal protection to minimize recurrent trauma, and physiotherapy [4]. Other therapeutic alternatives are repeated injections of Botox into the muscle [3, 5] or CT-guided PN blocks with anesthetics and steroids [4].

In cases with insufficient relief, surgical decompression can be offered. Entrapment occurs more commonly at the level of the ischial spine or at the entry of the Alcock canal [3, 8], and although different surgical approaches are described in the literature [4, 5, 8, 9] we prefer the laparoscopic transperitoneal approach due to minimal invasiveness, low morbidity, good access to the PN anatomic course and successful results [2, 9].

We describe a peculiar symptom not previously reported as associated with PNE, highlighting the wide range of symptoms that can arise from this condition. We argue that pain is not necessarily the main symptom in PNE, and that it must be acknowledged as a functional condition in order to properly diagnose and treat patients.

CONFLICTS OF INTEREST

The authors declare no conflicts of interest.

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