

Actinic cystitis associated with urinary tract infection by *Candida glabrata* as a differential diagnosis of pudendal neuralgia. Case report

*Cistite actínica associada à infecção de trato urinário por *Candida glabrata* como diagnóstico diferencial de neuralgia do pudendo. Relato de caso*

Lidia Yanka Hoffmann¹, Alexandre Daronco¹, Eduardo Hildebrand Seyboth²

DOI 10.5935/2595-0118.20200032

ABSTRACT

BACKGROUND AND OBJECTIVES: Chronic pelvic pain is still a little-known syndrome with different etiological agents, high morbidity rate, with little information about its etiopathogenesis, which makes its treatment difficult, with symptoms that significantly impact the patient's quality of life. Among the conditions that lead to chronic pelvic pain, actinic cystitis stands out, a complication of pelvic radiotherapy. This pathology presents signs, symptoms, and complications similar to pudendal neuralgia. The objective of this report is to present one of the etiologies of chronic pelvic pain and its treatment.

CASE REPORT: Sixty-three-year-old male patient who sought medical care with a history of chronic pelvic pain attributed to pudendal neuralgia. The diagnosis was actinic cystitis, resulting from pelvic radiotherapy for prostate adenocarcinoma and urinary tract infection by *Candida glabrata*, an opportunistic fungal agent. The patient was treated with fluconazole with total control of the painful symptoms and significant improvement in the quality of life.

CONCLUSION: A case report with total pain control in a patient with an initial diagnosis of pudendal neuralgia, who after the clinical evaluation, imaging tests, cystoscopy, and lab tests was diagnosed with actinic cystitis associated to the urinary tract infection by *Candida glabrata*.

Keywords: *Candida glabrata*, Cystitis, Pelvic pain, Pudendal neuralgia.

RESUMO

JUSTIFICATIVA E OBJETIVOS: A dor pélvica crônica é uma síndrome ainda pouco conhecida, com diferentes agentes etiológicos, com alta morbidade, com poucas informações sobre sua etiopatogênese, dificultando seu tratamento, com sintomas que impactam de modo significativo a qualidade de vida do paciente. Entre as inúmeras causas está a cistite actínica, complicação da radioterapia pélvica, que apresenta sinais, sintomas e complicações semelhantes à neuralgia do pudendo. O objetivo deste relato foi apresentar uma das etiologias da dor pélvica crônica e seu tratamento.

RELATO DO CASO: Paciente do sexo masculino, 63 anos, que procurou o serviço médico com história de dor pélvica crônica atribuída à neuralgia do pudendo. Foi feito o diagnóstico de cistite actínica causada por radioterapia pélvica para tratamento de adenocarcinoma de próstata e infecção de trato urinário pelo agente fúngico oportunista *Candida glabrata*, e instituído tratamento com fluconazol com controle total da sintomatologia dolorosa e melhora acentuada da qualidade de vida.

CONCLUSÃO: Relato de caso com controle total da dor em paciente que apresentava diagnóstico inicial de neuralgia do pudendo, que após avaliação clínica, imaginológica, cistoscópica e laboratorial foi diagnosticado com cistite actínica associada à infecção de trato urinário por *Candida glabrata*.

Descritores: *Candida glabrata*, Cistite, Dor pélvica, Neuralgia do pudendo.

INTRODUCTION

Chronic pelvic pain (CPP) is a disorder that affects about 4% of the population with chronic pain. It is a poorly diagnosed, complex syndrome, with many factors that can make diagnosis and treatment difficult. It is common for the patient to go through several professionals and, in most cases, remain without a precise diagnosis, considering the clinical condition and the multiplicity of etiologies. It is estimated that the average time between the onset of symptoms and diagnosis is around five and a half years^{1,2}.

The causes can be mechanical trauma of any etiology, immunological processes, infections, and/or inflammatory processes^{2,3}, so actinic cystitis is one of the causes of CPP⁴. Other causes include vulvodynia, psychosomatic disorders, prostatitis, urinary tract infection, pelvic inflammatory disease, and sexually transmitted infections⁵.

Lidia Yanka Hoffmann – <https://orcid.org/0000-0003-2176-7342>;
Alexandre Daronco – <https://orcid.org/0000-0002-1117-8803>;
Eduardo Hildebrand Seyboth – <https://orcid.org/0000-0001-7181-4624>.

1. Centro Universitário da Fundação Assis Gurgacz, Faculdade de Medicina, Cascavel, PR, Brasil.
2. Hospital Marechal Cândido Rondon, Marechal Cândido Rondon, PR, Brasil.

Submitted on July 15, 2019.

Accepted for publication on March 19, 2020.

Conflict of interests: none – Sponsoring sources: none.

Correspondence to:

Avenida Brasil, 5102 – Pacaembu
85816-302 Cascavel, PR, Brasil.
E-mail: lidiayanka@hotmail.com

© Sociedade Brasileira para o Estudo da Dor

Actinic cystitis has a clinical spectrum characterized by a series of post-pelvic radiotherapy manifestations, such as hematuria, urination pain, urinary incontinence, hydronephrosis, decreased bladder storage capacity, and propensity for urinary infections^{4,6,7}. It is a radiation-induced late endothelial lesion, associated with perivascular fibrosis, which causes ischemia and terminal obliterative arteritis, which can produce acute and chronic symptoms, worsening the patient's quality of life, especially when associated with the development of urinary infections^{7,8}.

CASE REPORT

A sixty-three-year-old male patient, diabetic, who sought care due to severe chronic pain. He was referred by the neurology service with a presumed diagnosis of pudendal neuralgia.

The symptoms started two years ago, after pelvic radiotherapy to treat recurrent prostate cancer. He presented paresthesia in the genital region, 7/10 pain assessed by the verbal rating scale (VRS), located on the topography of the pudendal nerve, which used to get better when sitting on the toilet and worsened throughout the day in the orthostatic position, with an average intensity of 8 on the visual analog scale (VAS). He had urinary incontinence as a result of radical prostatectomy and pelvic radiotherapy, change in the appearance of urine during the period when the intensity of pain and discomfort increased.

The therapeutic planning respected what is recommended, maintaining gabapentin and amitriptyline, in combination with prednisone and tramadol for a short period, obtaining good pain control, but maintaining the urinary complaint.

In the urine culture with the material collected in the bladder tube, there was the growth of *Candida glabrata*, but no sensitivity test to antifungals was performed. The Magnetic resonance imaging of the pelvis and the cystoscopy showed an imaging condition compatible with radiation cystitis.

The patient was referred to the infectious disease specialist who started antifungal therapy with fluconazole, with significant improvement in pain complaints and with zero pain intensity by both VRS and VAS, and absence of discomfort. The vesical delay probe was maintained because although possible ways of treating urinary incontinence have been proposed, and even being informed about the risks and benefits, he chose for the vesical delay probe. Throughout the follow-up, the pain complaints were entirely resolved, and all the drugs prescribed for the control of chronic pain were suspended, and the patient is still in outpatient follow-up with no complaints. This case study was approved by the Ethics Committee of the Centro Universitário da Fundação Assis Gurgacz (5219) and submitted to the National Research Ethics Commission - 11615119.4.0000.5219 CAAE, obtaining the formal authorization of the patient by signing the Free and Informed Consent Term (FICT).

DISCUSSION

Many patients with CPP have neuropathic pain in the pudendal nerve distribution; that is, located in the vulva, vagina, clitoris, perineum, and rectum in women; and in men, in the

glans, penis, scrotal pouch, except for the testicles, perineum, and rectum.

A small percentage of patients can have side symptoms, out of the area specifically determined by the pudendal nerve, since this region has a plexus-type innervation, so it may have several tributaries, causing manifestations in atypical topographies. Such symptoms can be a pain in the lumbar region, dyspareunia, altered urinary frequency, and urinary incontinence¹⁻³, hyperalgesia, and allodynia can add to the condition^{2,3}.

The CPP also has actinic cystitis as its etiology, which comprises a series of manifestations also present in other causes of CPP, including hematuria, urination pain, chronic pelvic pain, urinary incontinence, hydronephrosis and decreased bladder storage capacity, which in itself, are unspecific^{2,5,8}.

The diagnosis is made by the symptoms in patients undergoing pelvic radiotherapy, associated with imaging tests and cystoscopy, showing telangiectasis with friable erythematous mucosa, erythema, edema, reduced bladder capacity, fistulas or fibrosis⁹. In addition, tests such as ultrasound of the lower urinary tract, computerized tomography, and nuclear magnetic resonance can also provide the diagnosis of radiation cystitis^{6,10}.

In the present study, the patient had symptoms compatible with such conditions besides the presence of changes in the urine aspect, and *Candida glabrata* in the urine culture. *Candida glabrata* is a pathogenic agent in humans. This opportunistic fungus accounts for up to 29% of total bloodstream infections, in addition to causing infections of the urinary tract and vagina^{11,12}, recognized as important causes of morbidity and mortality, especially in immunosuppressed patients¹¹⁻¹³. Some of the risk factors for urinary tract infections by *Candida* species are immunosuppression, diabetes mellitus, prolonged hospitalization, urinary catheter, use of broad-spectrum antibiotics, female gender, and age over 65^{11,12}. Fluconazole is the drug of choice for *Candida glabrata*. However, in case of resistance to the drug, intravenous or intravesical amphotericin can be used^{11,12}.

CONCLUSION

A case report with total pain control in a patient who had an initial diagnosis of pudendal neuralgia, who after clinical, imaging, cystoscopic, and laboratory evaluation was diagnosed with actinic cystitis associated with infection by *Candida glabrata*.

REFERENCES

1. Pereira A, Pérez-Medina T, Rodríguez-Tapia A, Rutherford S, Millan I, Iglesias E, et al. Chronic perineal pain: analyses of prognostic factors in pudendal neuralgia. *Clin J Pain*. 2014;30(7):577-82.
2. Hibner M, Desai N, Robertson LJ, Nour M. Pudendal neuralgia. *J Minim Invasive Gynecol*. 2010;17(2):148-53.
3. Pérez-López FR, Hita-Contreras F. Management of pudendal neuralgia. *Climacteric*. 2014;17(6):654-6.
4. Mangano MS, De Gobbi A, Ciaccia M, Lamon C, Beniamin F, Maccatrozzo L. Actinic cystitis: causes, treatment and experience of a single centre in the last five years. *Urologia*. 2018;85(1):25-8.
5. Antolak SJ, Antolak CM. Chronic pelvic pain: neurogenic or non-neurogenic? Warm detection threshold testing supports a diagnosis of pudendal neuropathy. *Pain Physician*. 2018;21(2):E125-135.
6. Thompson A, Adamson A, Bahl A, Borwell J, Dodds D, Heath C, et al. Guidelines for the diagnosis, prevention and management of chemical-and radiation-induced cystitis. *J Clin Urol*. 2014;7(1):25-35.

7. Pascoe C, Duncan C, Lamb BW, Davis NF, Lynch TH, Murphy DG, et al. Current management of radiation cystitis: a review and practical guide to clinical management. *BJU Int.* 2019;123(4):585-94.
8. Sommariva ML, Sandri SD, Guerrer CS. [Treatment of acute iatrogenic cystitis secondary to bladder chemo-immuno-instillation or pelvic radiotherapy]. *Urologia.* 2010;77(3):187-92.
9. Smit SG, Heyns CF. Management of radiation cystitis. *Nat Rev Urol.* 2010;7(4):206-14.
10. Browne C, Davis NF, Mac Craith E, Lennon GM, Mulvin DW, Quinlan DM, et al. A narrative review on the pathophysiology and management for radiation cystitis. *Adv Urol.* 2015;2015:346812.
11. Pappas PG, Kauffman CA, Andes DR, Clancy CJ, Marr KA, Ostrosky-Zeichner L, et al. Clinical practice guideline for the management of candidiasis: 2016 update by the Infectious Diseases Society of America. *Clin Infect Dis.* 2015;62(4):e1-50.
12. Charlier C, El Sissy C, Bachelier-Bassi S, Scemla A, Quesne G, Sitterlé E, et al. Acquired flucytosine resistance during combination therapy with caspofungin and flucytosine for *Candida glabrata* cystitis. *Antimicrob Agents Chemother.* 2016;60(1):662-5.
13. Jiménez-Guerra G, Casanovas Moreno-Torres I, Gutiérrez-Soto M, Vazquez-Alonso F, Sorlózano-Puerto A, Navarro-Marí JM, et al. [Inpatient candiduria: etiology, susceptibility to antifungal drugs and risk factors]. *Rev Esp Quimioter.* 2018;31(4):323-8.

