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CASE REPORT

Urethral Carcinoma in Patients with Urethral Stricture: A Case Report

Zakaria Bakkali Issaoui, Youssef Kharbach, Abdelhak Khallouk
Urology department, Tangier University Hospital
Faculty of Medicine and Pharmacy, Abdelmalek Essaâdi University, Tangier, Morocco

ABSTRACT
Primary cancer of the urethra is rare, and its clinical presentation is nonspecific and urethral stricture is one of its risk factors. The authors report the case of a 65-year-old male patient with a history of recurrent urethral stricture for which he opted for a suprapubic catheter. He developed a perineal phlegmon that revealed a primary urethral carcinoma. This case shows unusual symptoms that should lead to suspect urethral carcinoma in a patient with urethral stricture. This will allow avoiding late diagnosis caused by misleading signs.

KEYWORDS: Urethral Neoplasms; Urethral Stricture; Squamous Cell Carcinoma; Urethral Carcinoma; Urethra.

Correspondence: Zakaria Bakkali Issaoui, Urology department, Tangier University Hospital, Faculty of Medicine and Pharmacy, Abdelmalek Essaâdi University, Tangier, Morocco. Email: zakariaissaoui753@gmail.com

INTRODUCTION

Primary cancer of the urethra is rare, and it accounts for less than 1% of all malignancies [1]. The prognosis of this disease is often unfavorable [2], and its clinical presentation is nonspecific and often mimics other conditions. Pathology depends on the portion of the urethra affected, and treatment options are indicated based on anatomical factors and the stage of the tumor.

The authors report the case of primary cancer of the urethra revealed by perineal phlegmon in a patient with a history of urethral stricture. The loco-regional extension of the tumor made any surgical intervention illusory. Chemoradiotherapy has recently given hope for the management of these tumors [2].

CASE REPORT

Mr. A.C is a 65-year-old male patient with a history of recurrent and poorly treated acute urethritis. He developed recurrent urethral stricture, for which he benefited from endoscopic urethrotomy on four occasions. The patient opted for a suprapubic catheter in situ for three years because of precocious recurrences of his stricture. He reported a 1-year history of an insidious appearance of a perineal induration for which he did not consult any medical doctor.

The patient's symptom was aggravated by the appearance of inflammation with swelling of the indurated lesion, which led to the formation of a perineal phlegmon with multiple fistulas (Figure 1). He had no anal disorders or rectal bleeding. Physical examination found a subcutaneous infection in the perineoscrotal area with bilateral palpable inguinal lymphadenopathy. Digital rectal examination found an average-sized prostate (about 50g) which was problematic inconsistency. We admitted the patient to the operating room for debridement. Per operative, the finding was an indurated tumor (about 5 cm in size) on the bulbo-membranous urethra with necrotic foci, for which we performed a biopsy. We could not do a cystoscopy because the urethral stricture was complete. The persistence of these lesions marked the evolution despite appropriate treatment.

Figure 1: Perineal phlegmon with multiple fistulas

Histopathological examination revealed a well-differentiated squamous cell carcinoma. We performed an abdominopelvic MRI as part of the tumor staging, which showed a large perineal mass involving the prostate, the different portions of the urethra, the scrotal area, and in direct contact with the anal canal. Multiple bilateral inguinal lymphadenopathies and vertebral metastasis were also found [figures 2, 3, and 4].

Our treatment choice was to perform chemoradiotherapy, but the patient died from massive pulmonary embolism before the start of treatment.

We present our findings after analyzing our clinical data and reviewing relevant published articles in the literature. We performed our bibliographic search on different databases: PubMed, ScienceDirect using the following keywords and MeSH: "Urethral neoplasms" and "Carcinoma, therapy." Afterward, a complimentary search on Google Scholar was made. Written informed consent was obtained from the patient for publication of this case report and all accompanying images.

Figure 2 : T2 sagittal sequence showing a voluminous perineal process in heterosignal encompassing prostate and ureter

Figure 3 : T2 axial sequence showing the voluminous process that comes into contact with the anal canal with loss of the separation border by location

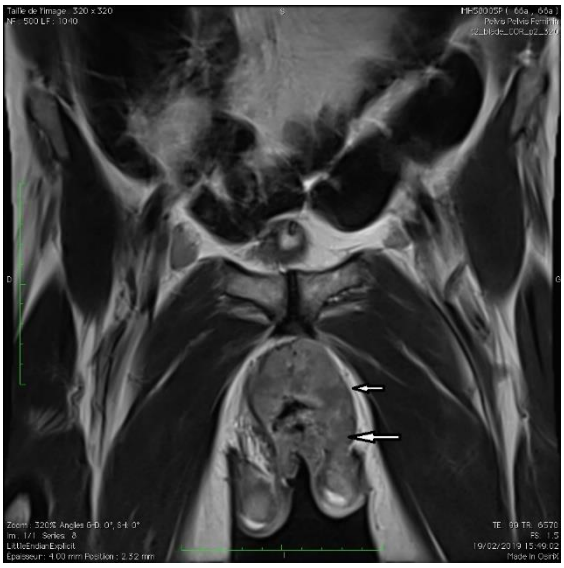

Figure 4: Coronal T2 sequence showing extension to the left purse

DISCUSSION

Primary carcinoma of the urethra is rare and aggressive, representing less than 1% of tumors of the urinary tract in humans [1,3], with limited data guiding treatment [3,4]. The average age of onset is 60 years [2], and it is predominant in women with a sex ratio of 3: 1 [2]. Several risk factors have been reported, including urethral stricture, chronic irritation, external radiotherapy, and urethritis secondary to sexually transmitted infections, including HPV [2,3,5], especially HPV 16 and HPV 18 [6].

Tumors that develop in the proximal urethra are often urothelial carcinomas or adenocarcinomas, whereas tumors of the distal urethra are mostly squamous cell carcinomas [6].

Squamous cell carcinoma subtype accounts for 68% of cases in men and 44.9% in women [7]. In men, the bulbo-membranous urethra, as in our patient, is involved in 2/3 of the cases [2].

Squamous cell carcinoma of the urethra occurs early with nonspecific signs; haematuria or urethrorrhagia [5]. When it becomes locally advanced, as in our patient, we have a palpable mass along the urethra, dysuria, pelvic pain, urethrocutaneous fistula, or abscess [5].

These nonspecific symptoms are often mistaken to be caused by a urethral stricture or a prostatic disease in men, hence the late diagnosis that usually occurs. Therefore, it is necessary to perform additional investigations in the presence of unusual events in known patients with urethral strictures, such as the appearance of genital swelling, rapid recurrence of symptoms after treatment, fistula, or a rapid worsening of symptoms dysuria despite intervention [2,8]. Other constitutional symptoms may be helpful to lead to the diagnosis, such as weakness, fever, anorexia, night sweats, or cachexia [8]. History of immunosuppression and family history of cancer should raise suspicion for possible urethral tumors [8]. Unfortunately, our patient did not consult earlier because he believed his symptoms were related to his urethral stricture.

The physical examination must look for an indurated mass and include a digital rectal examination [5]. Assessment of lymph node areas will look for the presence of inguinal lymphadenopathy.

Urine cytology has a low sensitivity (55 to 59%) [5]. Diagnosis of this disease is based on urethrocystoscopy with biopsy of suspicious lesions [5], especially in patients with gross or microscopic haematuria [8].

MRI is typically the imaging modality of choice in evaluating patients with urethral cancer [8]. It also plays an essential role in locoregional tumor staging. It provides information on the tumor site and the invaded surrounding structures [6], with very high accuracy of 93 % for clinical nodal staging to predict pathological lymph node involvement [9]. However, image interpretation requires particular expertise [8]. The tumor staging will also include a thoraco-abdominopelvic CT scan for possible metastases [5].

Tumour stage, tumor size, patient age, and lymph node involvement are the most important prognostic factors for survival without recurrence [9].

The therapeutic options depend on anatomical factors and tumor stage [3,4].

Schematically: Radical surgery is proposed for tumors of the bulbo-membranous urethra, which could be cystoprostatectomy or even emasculation [2,3,5]. A less aggressive approach is proposed for distal tumors consisting of conservative surgery (segmental urethrectomy) combined with chemoradiotherapy [3]. In locally advanced cancers (≥cT3 or cN+), cisplatin-based neoadjuvant chemotherapy is indicated before surgery [5]. Chemoradiotherapy is shown in inoperable patients as in our patients [5]. The prognosis is better for tumors of the anterior than tumors of the posterior urethra [2].

CONCLUSIONS

The rarity of primary carcinoma of the urethra explains the challenges related to the disease. A more thorough investigation should be performed when unusual events occur in known patients with urethral stricture or if they persist despite appropriate treatment. Our case perfectly illustrates the problems related to the late diagnosis caused by misleading and nonspecific symptoms.

AUTHORS' CONTRIBUTIONS

All the authors have actively participated in the redaction, the revision of the manuscript, and provided approval for this final revised version.

INFORMED CONSENT

Written informed consent was obtained from the patient.

CONFLICT OF INTEREST

Authors have no conflicts of interest to declare.

REFERENCES

[1] Gatta G, van der Zwan JM, Casali PG, Siesling S, Dei Tos AP, Kunkler I, et al. Rare cancers are not so rare: the rare cancer burden in Europe. Eur J Cancer. 2011 Nov; 47(17):2493-511. DOI. 10.1016/j.ejca.2011.08.008

[2] Ghorbel J, Hafsia G, Derouiche A, Jrad A, Chebil M. Carcinome épidermoïde de l'urètre masculin révélé par une rupture spontanée de l'urètre. Can Urol Assoc J. 2011 Jun; 5(3):E36-9. DOI : 10.5489/cuaj.09174

[3] Lucarelli G, Spilotros M, Vavallo A, Palazzo S, Miacola C, Forte S, et al. A challenging surgical approach to locally advanced primary urethral carcinoma: a case report and literature review. Medicine. 2016 May; 95(19):e3642. DOI: 10.1097/md.0000000000003642

[4] Son CH, Liauw SL, Hasan Y, Solanki AA. Optimizing the role of surgery and radiation therapy in urethral cancer based on histology and disease extent. Int J Radiat Oncol Biol Phys. 2018 Oct 1; 102(2):304-313. DOI: 10.1016/j.ijrobp.2018.06.007

[5] Gakis G, Witjes JA, Compérat E, Cowan NC, De Santis M, Lebret T. EAU Guidelines on Primary urethral carcinoma.. Eur Urol. 2013 Nov;64(5):823-30. DOI: 10.1016/j.eururo.2013.03.044

Zhang M, Adeniran AJ, Vikram R, Tamboli P, Pettaway C, Bondaruk J, et al. Carcinoma of the urethra. Hum Pathol. 2018 Feb ; 72:35-44. DOI : 10.1016/j.humpath.2017.08.006

[6] Iborra F, Rigaud J, Bastide C, Mottet N, les membres du sous-comité organes génitaux externes du comité de cancérologie de l'AFU. [Treatment of primary urethral carcinoma. Guidelines from the French Urological Association. Cancer committee]. Prog Urol. 2009 Mar; 19(3):170-5. DOI: 10.1016/j.purol.2008.12.003

[7] Grivas PD, Davenport M, Montie JE, Kunju LP, Feng F, Weizer AZ. Urethral cancer. Hematol Oncol Clin North Am. 2012 Dec; 26(6):1291-314. DOI: 10.1016/j.hoc.2012.08.006

[8] Gakis G, Morgan TM, Efstathiou JA, Keegan KA, Mischinger J, Todenhoefer T, et al. Prognostic factors and outcomes in primary urethral cancer: results from the international collaboration on primary urethral carcinoma. World J Urol. 2016 Jan; 34(1):97-103. DOI: 10.1007/s00345-015-1583-7

[9] Savoie PH, Fléchon A, Morel-Journel N, Murez T, Ferretti L, Camparo P, et al. [French ccAFU guidelines – Update 2018 – 2020 : Penile cancer]. DOI: 10.1016/j.purol.2019.01.008