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(CASE REPORT)



Urothelial carcinoma of the urethra: A new case and review of the literature

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Abstract

The urothelial carcinoma of the male urethra is a rare entity. This study reports a case of the urethra's urothelial carcinoma in a 71-year-old man who sought medical attention for an ulcerative-burgeoning swelling of the penis, centered on the meatus region (annex 1, 2).

The pelvic MRI performed had revealed a well-limited tissular ulcerative-burgeoning structure localized at the same level as the glans, with irregular contours. Its' histological examination has confirmed a urothelial carcinoma of the urethra.

The patient refused any additional treatment, the lymphadenectomy included. He then presented a local recurrence 4 months later.

Keywords: Urothelial carcinoma; Lymphadenectomy; Urethra

1. Introduction

Primary tumors of the urethra are rare.

They represent fewer than 1% of all the urinary tract's malignant tumors, regardless of their histological types.

Because the symptoms not being specific enough, the diagnosis is more often than not late or established by fortuitous discovery, which worsens the prognosis.

Our study aims at underlining this rare pathology's diagnostic and therapeutic difficulties, and at highlighting after a review of the literature, its' epidemiologic, diagnostic, and therapeutic aspects.

2. Case

Mr. GM is 71 years old man, who visited the urology department of the university hospital HASSAN II of FEZ for an ulcerative-burgeoning lesion localized in the meatus of the glans, that has been growing for the previous 6 months and has been resistant to medical treatment.

The patient has a history of high-risk prostatic adenocarcinoma staged Gleason 8 (4+4), which had been treated with radio-hormonal therapy, along with hepatitis C and post-radiation rectitis. His current t-PSA is at 0.07ng/ml.

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The clinical examination has revealed, a 1cm long, painless ulcerating mass of the glans, fixed to the deep plane, with no induration localized in the penile urethra and the corpus spongiosum.

2.1. Eternal genitalia examination

- The testicles are in place.
- The urethral meatus is invaded by the process.
- The lymph node areas are free, especially the inguinal ones
- Rectal examination: prostate of 30 grams firm with mobile bladder base.
- Note that the patient does not present signs of a lower urinary tract infection.

The pelvic MRI showcased a well-limited tissular, ulcerative-burgeoning, structure localized at the same level as the glans, with irregular contours. It was in hypo signal with T1 with a heterogenous contrast enhancement.

2.2. Relations and extensions

- Posteriorly: it invades the distal part of the urethra and the corpus spongiosum.
- Above and anteriorly: it remains distant from the corpora cavernosa.
- Laterally: bilateral, infra-centimetric, inguinal lymphatic formations

In front of this structure, a biopsy was performed and later revealed a high-grade pT1 urothelial carcinoma.

A ureteral-cystoscopy was performed and showed inflammatory lesions in the bladder's fundus, whose histological examination did not find any sign of malignancy.

And as an extension assessment, a thoraco-abdomino-pelvic scan was executed, showing no distant secondary lesion other than some infra-centimetric bilateral inguinal lymph node formations.

Finally, as a therapeutic conduct, a glandulectomy was carried out. The histological examination of its' product returned in favor of a poorly differentiated urothelial carcinoma whose nature was determined by an immunohistochemical study (P16 and CK5/6), and that was classified pT1 grade III with resection limits at 0.5 cm from the nearest peripheral resection and 0.4 cm from the deep limits, therefore pT1G3 N0 M0.

From then on, a lymphadenectomy was offered to the patient, which he declined.





Figure 1 and 2 An ulcerative swelling of the penis centered on the meatus

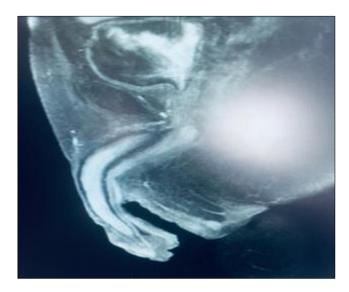


Figure 3 " hyposignal T1"

3. Discussion

With nearly 1500 cases listed in the literature since their first description by Thiaudière in 1834, primary urethral tumors are rare and represent, all histological types combined, fewer than 1% of malignant urinary tract tumors (1). Their histological nature is dominated by squamous cell carcinomas, which represent 70 to 90% of all urethral cancers (2). Urothelial carcinoma is the second histological type encountered (10 to 15%) and its primitive urethral location remains exceptional since it usually accompanies an invasion of the entire urinary excretory tract.

This case involving primary urothelial carcinomas is the first of its kind in the urology department of HASSAN II's University hospital.

Like many other malignant tumors of the urethra, urothelial carcinomas of the urethra affect subjects aged from 50 to 80 years old, but cases within the younger population have also been reported in the literature (4)

The etiopathogenesis of urethral carcinomas of the urethra is still not fully uncovered and no etiological factors have been identified with certainty. The main risk factors incriminated are recurrent infectious urethritis, post-infectious inflammatory stenotic lesions, post-traumatic inflammatory stenotic lesions, and the secondary/ synchronous localization of urethral carcinoma of the bladder or upper excretory tract (2,5,6). Urothelial carcinomas most frequently develop in the bulbar urethra, and rarely in the penile urethra and glans. (1,2,4,7)

Clinically, the symptoms are not specific and usually showcase a not-so-important narrowing of the urethra, which can delay the diagnosis (1.3). There are generally signs of low urinary obstruction, which can go from dysuria to acute urinary retention. These signs may or may not be associated with bloody discharge, pain in the perineum or penis, and genital disorders (2.8) such as painful erections or even an ulcerating mass.

In some cases, the discovery can be made during some therapeutic maneuvers such as the dilatation of urethral stricture or, in the most advanced forms, after the emergence of infectious complications (abscess, fistulas...).

However, the existence of indurated fistulous orifices giving way to a bloody discharge has a real semiological value, along with the presence of hard, fixed inguinal adenopathies, both of which were not found in our patient.

Urethrocystoscopy is the gold standard for tumor visualization and biopsy [9]. It also allows the visualization of the bladder and the elimination of any associated bladder tumor, which was the purpose of the urethral-cystoscopy we performed. [7]. The anatomopathological examination of the surgical biopsy allowed the diagnosis to be confirmed.

The histological characteristics depend on the tumor's site. Squamous cell carcinomas are the most common histological type, most often growing in the distal portion of the urethra. Urothelial carcinomas grow in the proximal part of the urethra and the bladder-neck region [3,4]. The rarer adenocarcinomas appear to develop from an existing para-urethral

diverticulum and often originate from the peri-urethral ducts [4]. Some neuro-endocrine histological types are extremely rare and of appalling prognoses such as malignant melanoma and small cell carcinoma [10]. They are diagnosed by immunohistochemistry (pan-cytokeratin, cytokeratin 20, and chromogranin) [10].

The extension assessment included an abdominopelvic CT scan (which revealed the presence of lymph node metastases), and magnetic resonance imaging (MRI).

The MRI provides an excellent image of the urethral wall and allows us to study the extension to the corpus spongiosum and the prostatic capsule, which was necessary in this case, as a way to determine the glandulectomy's limits.

The therapeutic management of primary urethral cancer depends essentially on its' stage and location. It involves surgery (partial or total amputation with perineal urethrostomy), radiotherapy / curative therapy, and chemotherapy [11] [15].

As for this case, before a distal localization and with the corpora cavernosa not being invaded a conservative surgery was proposed (glandulectomy). [Annexe 7]

Nevertheless, with the presence of bilateral inguinal adenopathies, we proposed a lymphadenectomy, which was again, refused by the patient. The management of our patient, in our social context, makes the treatment approximate and the prognosis pejorative [15]. (figure of the dissections' recommendations).

Indeed, the prognosis is correlated with the stage and grade [13]. Similarly, the prognosis depends on the location of the tumor and is poor when it is located in the bulb-membranous urethra since the diagnosis is more often than not done later. The prognosis, in this case, is also related to the treatment administered, which was unfortunately incomplete. Our patient had, hence, a local tumor recurrence 1 month later.

Epidemiologically, five-year survival is estimated at 36% for high-grade tumors and 83% for low-grade tumors [13,14]. For Dalbagni and al [14], the five-year survival is estimated at 69 and 26%, respectively for lesions of the anterior urethra and the bulbs-membranous urethra. The majority of deaths are related to local complications (sepsis and hemorrhage), recurrences, and lymph node metastasis. This prognosis can be improved by early diagnosis allowing non-mutilating conservative surgery.

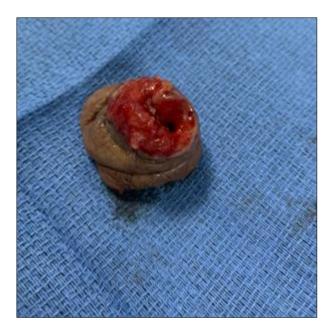


Figure 4 Surgical specimen of the glandulectomy



Figure 5 1-month post-surgery 1 month

4. Conclusion

Primary urothelial carcinoma of the urethra is very rare. It usually occurs within a population of men aged over 60 years old. Its symptomatology is no specific, hence the delay in the diagnosis. Systematic histopathological examination of urethral biopsies or surgical specimens confirms the diagnostic.

Primary tumors of the urethra remain rare and have a very serious prognosis despite the various treatments proposed. Their rarity makes it difficult today to evaluate the different therapeutic means.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of ethical approval

The present research work does not contain any studies performed on animals/humans subjects by any of the authors'.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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