Fait clinique

Squamous Cell Carcinoma of the Male Urethra Revealed by Urethro-scrotal Fistula: a Case Report.

Carcinome épidermoïde de l’urètre masculin révélé par une fistule urétro-scrotale : à propos d’un cas.

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ABSTRACT

Primary cancer of the male urethra is rare and of unknown etiology. The average age of onset is 60 years. It is mostly located on the bulbo-membranous urethra. We report a case of primary squamous cell carcinoma of the membranous urethra, discovered when exploring an urethro-scrotal fistula with chronic urinary retention. An urethrocystogram was suggestive of filling defect in the membranous urethra associated to urethro-scrotal fistula. Doppler ultrasound of the urethra confirmed the presence of an endo-urethral mass. Histology of the urethral partial resection concluded of squamous cell carcinoma of the membranous urethra. In the light of this case, we discuss the clinical features and difficulties in early diagnosis of these cancers in low-income countries.

RÉSUMÉ


1. Introduction

Urethral tumors account for less than 1 percent of the tumors of the urinary system [1]. Squamous cell carcinoma is the most frequent histological type (80 percent of cases) of primary cancer of the male urethra [2]. It is mostly located on the membranous part of the posterior urethra. Clinical signs vary, while the enabling factors are numerous [3]. Tumors of the posterior urethra are subject to poor prognosis, as the diagnosis usually performed when they have spread locally. We report a
case revealed by spontaneous trans-tumoral urethrocrotal fistula with chronic urinary retention.

2. Observation

M. T.H. of 65-year-old was referred to our Radiology Service for suspicion of urethral stenosis associated with urethrocrotal fistula. The history of the disease revealed dysuria with a progressive decline in urine stream for five months, followed by both a painful swelling of the right side of the scrotum and purulent scrotal outflow with urinary leaks through the scrotal wound during micturition.

The patient’s past history revealed chronic alcoholism and post-traumatic fracture of the right femoral neck aggravated by a shortening of the lower limb and chronic pains in the hips. Primary clinical examination showed that the patient was apparently in good health with normal temperature. Urological examination revealed a distended bladder, a large inflammatory swelling of the right scrotum with purulent wound. Digital rectal examination indicated a soft and normal-size prostate. Palpation of the flanks was unremarkable. We concluded of possible urethral stenosis with urethrocrotal fistula.

The initial management consisted of a bacteriological analysis of pus from the fistula, which revealed fosfomycin-sensitive E. Coli. Cytobacteriological examination of urine showed multi resistant Citrobacter freundii that was also sensitive to fosfomycin. Fasting blood sugar and creatininemia were normal. A transvesical catheter was performed, and adapted antibiotherapy was administered to the patient alongside with “seat-baths” for a total period of 15 days. At the end of this treatment, suppuration resolved but there remained a fistula that enabled urine leakages during micturition when the trans-vesical catheter was clamped.
Medical imaging procedures were requested. Retrograde and voiding ureterocystothrogram showed a millimetric annular stenosis (figure 1) associated with a swampy urethral lacuna of about 2 cm long anterior to the stenosis, and a right-side urethro-scrotal fistula communicating with the urethral lacuna (figure 1).

Ultrasound of the urinary system unveiled an intraluminal soft tissue mass of the post urethra-bulb corresponding to the lacuna observed using retrograde ureterocystothrogram. This mass was hyper echoic heterogeneous and ill-defined posteriorly, with poor Doppler spots (figure 2). Imaging and clinical findings were suggested of urethral cell carcinoma with urethro-scrotal fistula.

The patient undergo surgery. Surgical operation consisted of fistula excision and partial urethrectomy to remove the endo-urethral tumor and the annular stenosis, followed by perineal urethrostomy to open out the proximal urethra to the perineum in order to ensure micturition after removal of sub-pubic catheter.

Histopathology of the resected tumor revealed an invasive, well differentiated squamous cell carcinoma of 3 cm long axis size and around urethra-scrotal fistula track, with healthy resection margins.

As part of extension work-up, abdominal and pelvic ultrasound as well as a thoraco-abdomino-pelvic CT-scan were carried out. Both examinations were unremarkable. After 15 months of follow-up the patient is stable and no clinical alarm has been recorded.

3. Discussion
Primary cancer of the male urethra is rare [1]. It is mainly located in the bulb-membranous urethra (75 percent), and the most frequent histological type is the squamous cell carcinoma (80 percent of cases) [2]. The average age of onset is 60 years [4]. Urethral tumors account for less than 1 percent of the tumors of the urinary system [1]. The etiology of primary cancer of the male urethra remains unknown. Some enabling factors are being set forth, and have in common a chronic urethral irritation [3] like urethral stenosis found in 24 to 78 percent of urethral carcinomas; medical history of sexually transmissible genital infection found in 24 percent of cases; and history of urethral trauma in 7 percent of cases. Some authors indicate the role of HPV 16 and 18 (60 percent of female cases and 30 percent of male cases) [5, 6]. In this case, no enabling factor has been identified but the evidence of annular urethral stenosis maybe an after-effect of chronic infection.

About 65 to 75 percent of urethral tumors are located in the bulb-membranous urethra [1, 7]. They are much rare in the anterior urethra. In contrast, they are exceptionally located in the prostatic urethra. In the present case, we have a tumor of the bulb-membranous urethra.

Clinically, 94 percent of cases present symptoms, but of no specificities [8]. As the tumor is located in the anterior urethra, diagnosis is faster [9]. However, these symptoms may be wrongly attributed to trivial urethrostenosis, thus leading to late diagnosis. Obstructive symptoms are the most common (43 percent). Some 20 to 62 percent of cases present urethrorrhagia, while 20 percent of cases show irritative urinary symptoms. Hematuria leads to diagnosis in 17 percent of patients. Some 11 to 33 percent of cases present pelvi-perineal pains.

Clinical examination shows a perineal or urethral mass in 28 to 52 percent of cases [4, 10]. Any urethral fistula (10 percent) or perineal abscess (5 percent) in an elderly patient should be properly investigated. Like in our case, fistula accounts for 10 percent of revelation modes for urethral carcinoma [9]. This may be attributed to both the trans-tumoral fracture of the membranous urethra by the tumoral invasion of the urethral wall and to its weakening. Typically, urethro-scrotal fistula develops during an infection of the peri-urethral environment or of the Cowper glands, together with a stenosis-related urethral lesion favoring suppuration diffusion and fistula track formation. It may also complicate the surgical operation of the urethra. Ganglionic areas are affected in 30 percent of cases. Unlike penile carcinomas, 90 percent of palpable ganglionic areas are metastatic [4]. In this case, perineal abscess that secondarily fistulated to the skin was identified. The symptoms were initially considered trivial by the patient, but initial consultation showed it was infectious, as diagnosis was late. This is rare enough in the development of tumor and in an exceptional etiology of urethral rupture by both the tumoral invasion of the urethral wall and its weakening.

Diagnosis was confirmed following fistula lancing and post-operative discovery of tumor in the post-bulb urethra.

Urethrocystogram helps to evoke diagnosis when showing the characteristic images as it is the case for our
patient; that is, images of stenosis displaying extended, off-center tumoral stricture with irregular and uneven edges, all of which indicate the presence of burgeoning and ulcerated tumor whose infiltrating component narrows urethral lumen [11].

Clinico-radiological diagnosis is confirmed by histology. Cancers of the male urethra vary according to the original tissue: squamous cell carcinoma, urothelial carcinoma, and adrenocarcinoma account respectively for 68, 17.5 and 4.6 percent [12]. Cytological examination has a great diagnostic value when positive. Anatomo-pathological analysis of the tumoral exeresis part of our patient revealed invasive, well differentiated squamous cell carcinoma, which is the most frequent urethral cancer.

Cancer of the male urethra is a disease with locoregional malignancy which extends to erectile organs (corpus cavernosum or corpus spongiosum) or to regional lymph nodes. Inguinal lymph nodes are found in one third of cases [3]. Only 10 to 20 percent of cases present metastases [3]. We did not demonstrate any secondary localization in our case.

The type of treatment is mainly conditioned by tumoral topography, rather than by the histological type. For T1-2 lesions of the anterior urethra, the most used option is surgery [13]. However, treating primary lesion consists of an extended surgical resection, which depends on the location and stage of tumor. The surgical method selected for our patient consisted of partial uretrectomy to remove the tumor and the annular stenosis, coupled with perineal urethrostomy. This broad resection proved curative for the lesion, as confirmed by histology.

Prognosis of the lesions of the anterior urethra is better than that of the lesions of the bulbo-membranous urethra. Dalbagni et al [10] argue that survival after 5 years is respectively 69 percent and 26 percent for the lesions of the anterior urethra and those of the bulbo-membranous urethra. Tumors of the posterior urethra receive a late diagnosis because they are less accessible to investigations and therefore have a worse prognosis.

4. Conclusion
Primary cancer of the male urethra is a rare entity. Urethro-cutaneous fistula associated with irregular urethral lacuna is very suggestive. Urethrocytogram is crucial in early diagnosis. Any aged patient's urethral fistula should suggest diagnosis.

Competing interests
The authors declare that they have no competing interests.

5. References