African Journal of Urology
1110-5704

Vol. 15, No. 2, 2009
107-110

Case Report

Pseudotumoral Malacoplakia of the Bladder

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ABSTRACT

Malacoplakia is a rare inflammatory condition most often affecting the genitourinary system. We report the case of a 24-year-old man who presented with gross hematuria, nocturia, frequency, dysuria and considerable weight loss during the preceding three months. Digital rectal examination showed a solid pelvic mass. Ultrasonography and computed tomography showed calyceal dilatation on the right side and a solid bladder mass 10 cm in diameter suspicious of bladder cancer. Transurethral resection of the tumor was incomplete, due to the large volume of the bladder mass. Histological examination of the resected specimen revealed malacoplakia of the bladder. The patient was treated with fluoroquinolone and vitamin C. Follow-up at 3 months showed marked regression of the bladder mass and complete resolution of the calyceal dilatation.

Key Words: Malacoplakia, bladder, pseudotumor, antibiotics.

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Article Info : Date received: 25/5/2008 Date accepted (after revision): 14/1/2009

INTRODUCTION

First described in 1902 by Michaelis and Gutmann[1], malacoplakia is a rare, benign inflammatory disease mostly affecting the genitourinary system[2]. The diagnosis is exclusively based on histological examination and treatment consists of a combination of long-term antibiotic therapy, cholinergic agonists and/or ascorbic acid (vitamin C). We report a case of pseudotumoral malacoplakia of the bladder in a 24-year-old man which was clinically misdiagnosed as bladder malignancy.

CASE REPORT

A 24-year-old-man was admitted to our department for gross hematuria associated with nocturia, frequency and dysuria. The patient also complained of intermittent limping with psoitis pain and weight loss of 14 kg during the preceding 3 months. Digital rectal examination showed a solid pelvic mass. Ultrasonography demonstrated calyceal dilatation on the right side and a homolateral solid mass in the bladder, 10 cm in diameter. Computed tomography (CT) was suggestive of a localized bladder tumor (Fig. 1).

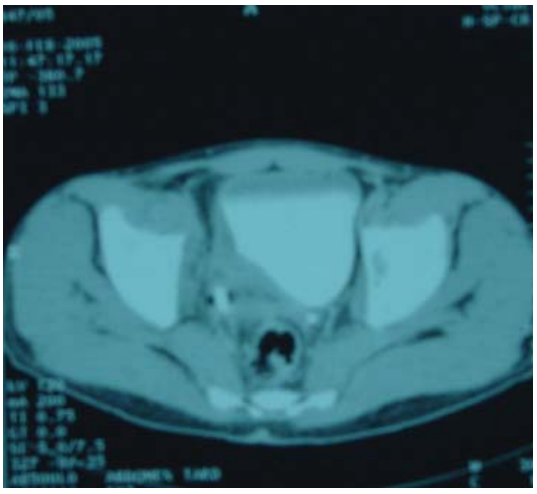

Fig.1: CT showing a solid mass in the right bladder wall.

Voiding cystourethrography revealed a large filling defect arising from the bladder

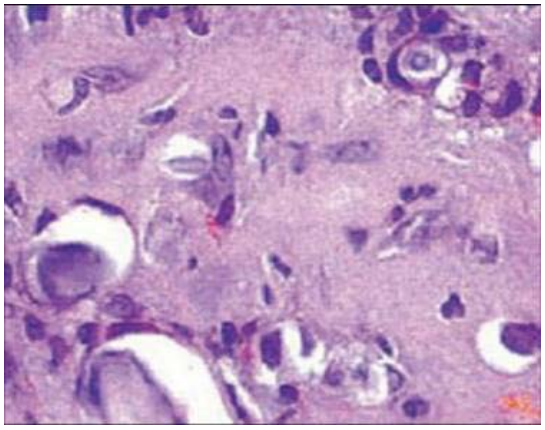

Fig. 2: Photomicrograph showing large macrophages with foamy eosinophilic cytoplasm (von Hansemann cells) and intracytoplasmic crystalline bodies (Michaelis-Gutmann bodies) (HE x 400).

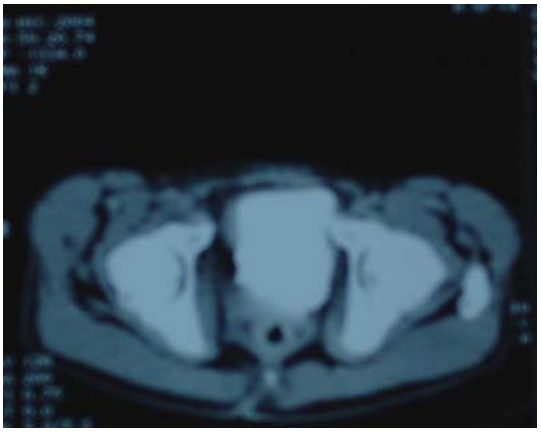

Fig. 3: Control CT 3 months after antibiotic therapy showing marked regression of the bladder mass.

wall and a grade-1 vesicoureteral reflux on the left. Urine culture revealed *E. coli* infection.

Based on these findings a malignant bladder tumor was diagnosed, and we decided to perform a transurethral resection of the bladder tumor (TURBT). Cystoscopic findings showed a yellow-white, soft mass with central umbilication. The bladder tumor could not be resected completely because of its large volume, which made it difficult to identify the bladder wall. We therefore decided to wait for the results of histological examination before completing resection of the residual tumor or performing total cystectomy. Surprisingly, the histological changes were consistent with malacoplakia of the bladder (Fig. 2).

The patient was then discharged and treated medically with fluoroquinolone and vitamin C. He remained asymptomatic, and CT done three months later revealed marked regression of the bladder mass and complete resolution of the calyceal dilatation (Fig. 3). The urine culture was negative on follow-up.

DISCUSSION

Malacoplakia has been reported in various sites (gastrointestinal and respiratory tract, retroperitoneum, skin, adrenal gland, tonsils, conjunctiva and pancreas)[3], but in the majority of cases (75%) it involves the urinary tract[2,4]. In the genitourinary form of the disease, the bladder is most commonly affected (70%), followed by isolated upper urinary tract lesions (15%) and combined upper and lower tract lesions (15%)[4]. It is more common in females (sex-ratio: 4:1) with a peak of incidence around 50 years of age[3,5]. Malacoplakia is very rare in children[6,7] and its incidence is probably underestimated in adults[8].

Tumoral malacoplakia of the bladder is an exceptional condition[9,10]. There is no imaging technique that can diagnose malacoplakia with certainty. Ultrasound may demonstrate renal dilatation or hyperechoic lesions, while the bladder may be small and contracted with soft tissue masses[10]. CT scan and magnetic resonance imaging (MRI) findings are also non-specific. Malignant bladder tumors represent the main differential diagnosis.

The diagnosis is exclusively histological after biopsy or TURBT. Malacoplakia is characterized by aggregates of large macrophages with foamy eosinophilic cytoplasm that are present at the sites of infection and exhibit numerous secondary lysosomes (von Hansemann cells). The fusion and calcification of these lysosomes result in the formation of intracytoplasmic crystalline bodies with a central hydroxyapatite core (pathognomonic Michaelis-Gutmann bodies) that give the macrophages a concentric target-like appearance[1,11].

The etiology is most likely an acquired defect in the bactericidal function of the macrophages that may result in incompletely digested bacterial fragments which undergo subsequent mineralization[2,12,13]. It is believed that the disease may be related to defective lysosomes and an abnormal microtubular assembly, similar to that seen in the Chediac-Higashi syndrome[12]. Microtubules are responsible for normal invagination and degranulation of lysosomes which are essential steps in the killing of bacteria by phagocytosis[13]. Alterations in the redox state with a reduction in the cGMP/cAMP ratio may be the main defect in malacoplakia[1]. Cholinergic agents enhance cGMP and beta glucuronidase, whereas vitamin C reduces cAMP. These agents can therefore be therapeutically effective in malacoplakia[1,3,13]. Malacoplakia has also been observed in association with immunocompromise, suggesting a disorder of the immune system[5]. Most of the cases of malacoplakia affecting the genitourinary system (90%) are associated with coliform infection, with *E. coli* being the most frequently isolated organism (72%)[1]. Various other possible causes of malacoplakia (prolonged treatment with systemic corticosteroids, tuberculosis, sarcoidosis, fungal or viral infection, and neoplasia) have been proposed[14]. We postulate that vesicoureteral reflux in our patient resulted in chronic bacteriuria with *E. coli.*

The association of malacoplakia with carcinoma or tuberculosis is classically described and can be observed in all organs, but specifically in the colorectal region.

Malacoplakia confined to the lower urinary tract can usually be treated medically. Persistent masses may require resection and/or the insertion of a ureteral stent if there is obstruction. In our patient resection allowed excluding the presence of bladder malignancy, reducing the volume of the bladder mass and clearing the right ureter. Antibiotics that achieve high intracellular levels such as quinolones, trimethoprim and rifampicin, have been shown to be most effective[1]. Quinolones are effective in 80% to 90% of cases. Trimethoprim and rifampicin are useful over prolonged periods. The addition of bethanecol chloride (Urecholine) and/or vitamin C may enhance phagocytic bactericidal activity[1,3,13,15]. Any immunosuppressive medications should be discontinued, if possible[5].

Monitoring of the patients should be extended because of the risk of recurrence. In this patient close follow-up and the maintenance of sterile urine have resulted in a marked regression of the vesical mass.

In conclusion, malacoplakia is a rare benign inflammatory condition which affects the genitourinary system in the majority of cases. Tumoral malacoplakia of the bladder is very rare and may be misdiagnosed as a malignant bladder tumor. Diagnosis is exclusively based on histology. Malacoplakia confined to the lower urinary tract is usually treated medically, but persistent or very large masses may require resection.

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