Inflammation and Infection

A Case Report of Xanthogranulomatous Cystitis

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ABSTRACT

Xanthogranulomatous cystitis (XC) is a chronic granulomatous inflammation with rare incidence. Herein, we report a 62-year-old female whose cystoscopic biopsy showed chronic inflammation accompanied with acute episode of inflammation. According to CT scan, pathology of the lesion could not rule out bladder carcinoma. Thus, partial cystectomy was done. But post-operatory pathological examination of the specimen was finally demonstrated Xanthogranulomatous cystitis. To the best of our knowledge, the present case of XC is the 28th to be reported in the world literature.

Introduction

Xanthogranulomatous cystitis (XC) is an unusual, benign, chronic inflammatory disease of the bladder. XC imitates malignant bladder tumor with unclear etiology, which was first described in 1932 by Wassiljew.1 Pathologically, XC is characterized by xanthoma cells (lipid-laden macrophages), multinucleated giant cells, cholesterol clefts, fibrosis and calcification. It has been reported in many sites, such as the kidney, gall bladder, colon, appendix, ovary, pancreas, salivary glands and endometrium.2 But it is extremely rarely discovered in bladder and usually misdiagnosed. Here we present a case of XC in which the patient's biopsy was first diagnosed as chronic inflammation.

Case presentation

A 62-year-old woman presented with pollakiuria, urgency, dysuria for 4 months. She denied having had any renal colic or painless hematuria and no history of urinary calculus. She had no significant past medical and drug abuse history. Laboratory examination brought out a normal hematological and biochemical profile. Urinalysis revealed 458.30 white blood cells and 36.40 red blood cells per microliter. Urine culture was negative.

Computerized tomography abdomen showed a protruding mass lesion (1.5 cm) in the anterior wall of the bladder. Cystoscopy-guided biopsy of the mass revealed chronic inflammation accompanied with acute episode of inflammation, partial mucosa was moderate atypical hyperplasia. Therefore, partial cystectomy was done. Epirubicin was perfused into bladder before and after operation. The specimen measured 4 cm in length and 4 cm in width. Pathological examination of the specimen was demonstrated Xanthogranulomatous cystitis. Histologically, biopsy of the mass was characterized by the presence of a dense infiltrate of foamy histiocytes, plasma cells, lymphocytes and multinucleated macrophages. Lymphoid follicle and urachal remnant were also detected in the lesion. Special immunohistochemical stains, including AAS stains, PAS stains, GARM stains and GMS stains were applied on suitable section (Fig. 1). None of the above stains was positive. The postoperative course was uneventful.

Discussion

XC is an extremely rare benign chronic inflammatory disease. To the best of our knowledge, about 28 cases have been reported in the world literature and majority of the reported cases are associated with urachal cyst or remnant. The major clinical symptoms were lower abdominal mass and cystitis like symptoms, such as frequency, urgency and dysuria. Of the 28 cases of Xanthogranulomatous cystitis in the literature, sixteen (57%) were associated with heterogeneous mass adherent to the anterior wall of the bladder.
and twelve of them (43%) were adhered to the dome of the bladder.3–5

The etiology of XC has not been clearly clarified. The probable etiologic factors which influence the development of XC are multitudinous, including urachal cyst or urachal adenoma, immunological disorders, chronic bacteria infection, an abnormal host response to a malignancy, abnormal lipid metabolism and lipid accumulation in macrophages.3 It is worth mentioning that Malakoplasia should be considered in the differential diagnosis. Malakoplakia, which is another rare form of chronic inflammatory granulomatous disease in the bladder usually occurring in adults with immunocompromised status or debilitating disease. It is diagnosed by the finding of foamy histiocytes with characteristic basophilic inclusions (Michaelis–Gutmann bodies). However, MG bodies were not found in the pathological specimen. Additionally, most of the reported cases present with lower abdominal mass and cystitis like symptoms, which is easy to confuse the other disease of bladder, particularly the malignant tumor. It is worth mentioning that a few of them were associated with urothelial carcinoma or prostatic adenocarcinoma3 (1 case). In this case (Fig. 2), the mass adherent to the anterior wall of the bladder was smooth, and the ulcer of the bladder tumor was not found. But we did not discover the inflammation changes such as congestion, edema. However, the lesion could not be excluded from the tumor with biopsy of the samples. So partial cystectomy was performed. Although a trial of long-term broad-spectrum antibiotics was suggested, conservative medical therapy is rarely employed. And the gold-standard curative treatment is surgical resection.

XC mimic malignancy macroscopically, which may be misdiagnosed as carcinoma before surgery. For the unclear lesions in the anterior wall of the bladder, especially to differentiate with urachal carcinoma. However, in the preoperative case, the rare XC lesions of the anterior wall of the bladder may be considered.

Conflict of interest

No conflict of interest.

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