

Case Report

Xanthogranulomatous cystitis imitating bladder neoplasm: a case report and review of literature

Ye Wang^{1*}, Xiang-Chun Han^{2*}, Li-Qiang Zheng³, Wen-Long Miao⁴

¹Department of Urinary Surgery, Tangshan Gongren Hospital, Tangshan, P. R. China; ²Department of Pathology, The First Affiliated Hospital to Hebei North University, Zhangjiakou, P. R. China; ³Department of Dermatology, 251 Hospital of PLA, Zhangjiakou, P. R. China; ⁴Department of Urinary Surgery, The First Affiliated Hospital to Hebei North University, Zhangjiakou, P. R. China. *Equal contributors.

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Abstract: Xanthogranulomatous cystitis is a rare, benign chronic inflammatory disorder with unclear etiology. Since its similar features to bladder neoplasm, it is extremely easy to be mistaken for malignant tumor. We herein reported a case of xanthogranulomatous cystitis in a 54-year-old male who presented with low abdominal pain and painless, total macrohematuria. Microscopy showed proliferation of large foam histiocytes which expressed CD68 and vimentin. Ki-67 showed a lower proliferation index (< 10%). Histopathology and immunohistochemical findings confirmed the diagnosis. This case highlighted the significance of recognizing this unusual lesion and differentiating it from its histological mimics by immunohistochemical staining.

Keywords: Xanthogranulomatous cystitis, neoplasm, CD68, vimentin, Ki-67

Introduction

Xanthogranulomatous cystitis (XC) is a rare, benign chronic inflammatory disorder of unclear etiology that was first described in 1932 [1]. Since its similar features to bladder neoplasm and it is extremely easy to be mistaken for malignant tumor both clinically and pathologically [2, 3]. To our knowledge, very few cases of XC have been reported in the literature [2, 4]. Herein we reported a case of XC in a 54-year-old male who presented with low abdominal pain and painless, total macrohematuria. Microscopy revealed proliferation of large foam histiocytes which expressed CD68 and vimentin. Histopathology and immunochemical staining confirmed the diagnosis. We highlighted significance of this unusual lesion, differentiated it from its histological mimics and reviewed the related literatures.

Case report

A 54-year-old male presented with low abdominal pain for half a month and painless, total macrohematuria intermittently for 3 days. His past and family histories were noncontributory.

He was otherwise healthy and the results of a physical examination were normal.

Laboratory studies revealed a normal hematological and biochemical profile. Urinalysis showed plenty of red blood cells per high power field. Urine culture was negative and urine cytology revealed no malignant cells. Due to our limited hospital conditions, CT examination of the urinary system could not be implemented.

Cystoscopy revealed a sessile, papillary and protruded lesion (3.0 cm x 2.0 cm) at the dome of the bladder. Biopsy of the mass suggested mild cystitis. Furthermore, the mass was presented with local perivesical invasion and the presence of bladder neoplasm could not be ruled out, so complete endoscopic resection was not done. The patient underwent surgical exploration. Frozen sections were taken during the surgery and the histopathologic results showed the mass was in relation with the visceral peritoneum and serous membrane of sigmoid with fibrotic adhesions. So the bladder was released from the mass and finally partial cystectomy was performed.

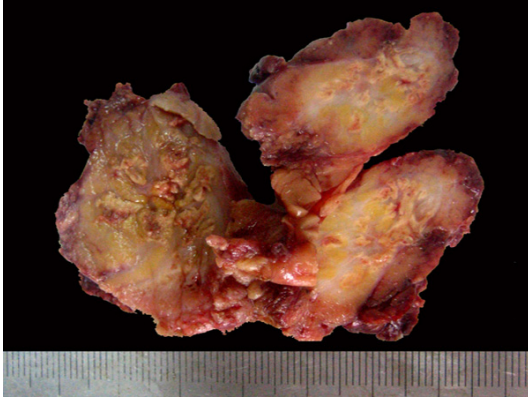


Figure 1. Gross findings reveal yellow changes with areas of necrosis.

Gross examination of the sessile mass indicated yellow changes with areas of necrosis (**Figure 1**). There was no anatomical relation between the XC lesion and the urachus.

Histological findings showed XC was characterized by foamy histiocytes, eosinophils and lymphocytes infiltration (**Figure 2A**). Special immunohistochemical stains, such as CD68, vimentin, ki-67 and CK (cytokeratin), were applied on appropriate sections. CD68 highlighted positivity (**Figure 2B**). Vimentin was also positive but CK negative (**Figure 2C**). Ki-67 showed a lower proliferation index ($< 10\%$) (**Figure 2D**). The postoperative course was uneventful. The patient remains asymptomatic at 6 months follow-up after treatment.

Discussion

Xanthogranulomatous inflammation is a type of chronic granulomatous inflammation histologically characterized by lipid-laden macrophages, multinucleated giant cells, cholesterol clefts, polymorphonuclear leukocytes, plasma cells and lymphocytes. Xanthogranulomatous lesions have been reported to involve many organs, including the gallbladder, pancreas, appendix, colon, ovary, endometrium, brain, and kidney, usually mimicking malignancy both clinically and microscopically [2-6]. XC is an extremely rare, affirmative chronic inflammatory disease with uncertain etiology, which was first described by Wassiljew in 1932 [1].

Up to now, more than twenty cases have been reported in PubMed literatures [4], with the fol-

lowing clinical characteristics: median age of 45 years; no obvious sexual predilection; the majority of lesions were located in the bladder dome (85.7%) and mostly associated with an urachal remnant (70.8%) [4].

The disease does not have specific clinical findings other than lower abdominal pain and cystitis like symptoms, umbilical discharge and occasional hematuria. Generally speaking, most of cases were localized mainly at the dome of the bladder or near the dome [4]. Occasionally, Yang et al reported an unusual case with a mass locating on the posterobasal wall of the bladder, which indicated an absence of relation with the urachus [7].

The etiology of XC is unclear. Proposed explanations suggest a chronic inflammatory process caused by mechanisms such as (a) immunological defect of the macrophage, (b) gram negative or anerobic bacteria such as in urinary tract infections or infection after tubal ligation [8], (c) chronic infection of the urachal diverticulum or cyst, (d) foreign material such as retained suture material [9], (e) local response to a bladder tumor [10, 11], (f) abnormal lipid metabolism and accumulation and (g) inflammatory bowel disease [12]. In the kidney, xanthogranulomatous inflammation almost always developed as a response to chronic, low-grade suppurative infection, often as a result of obstruction of flow of urine. Our case raised probably in a similar manner, in response to chronic infection without obvious symptoms.

The symptoms of XC are nonspecific and difficult to distinguish from other disorders of the bladder. Therefore, diagnosis is very difficult without tissue biopsy or without examining the tissues obtained from surgery. Even in the cases that we have examined, we suspected bladder tumor before the surgery but were able to diagnose the mass as XC when we discovered xanthoma cells with foamy cytoplasm with numerous infiltrations of inflammatory cells and an abundance of lipids in the tissue biopsy following the surgery [13].

Since medical treatment is ineffective, conservative management is rarely employed [4, 13]. The curative treatment of choice is surgical resection [13]. Localized disease may be amenable to simple tumor excision. However, it is sometimes difficult to distinguish between XC and XC accompanying carcinoma before opera-

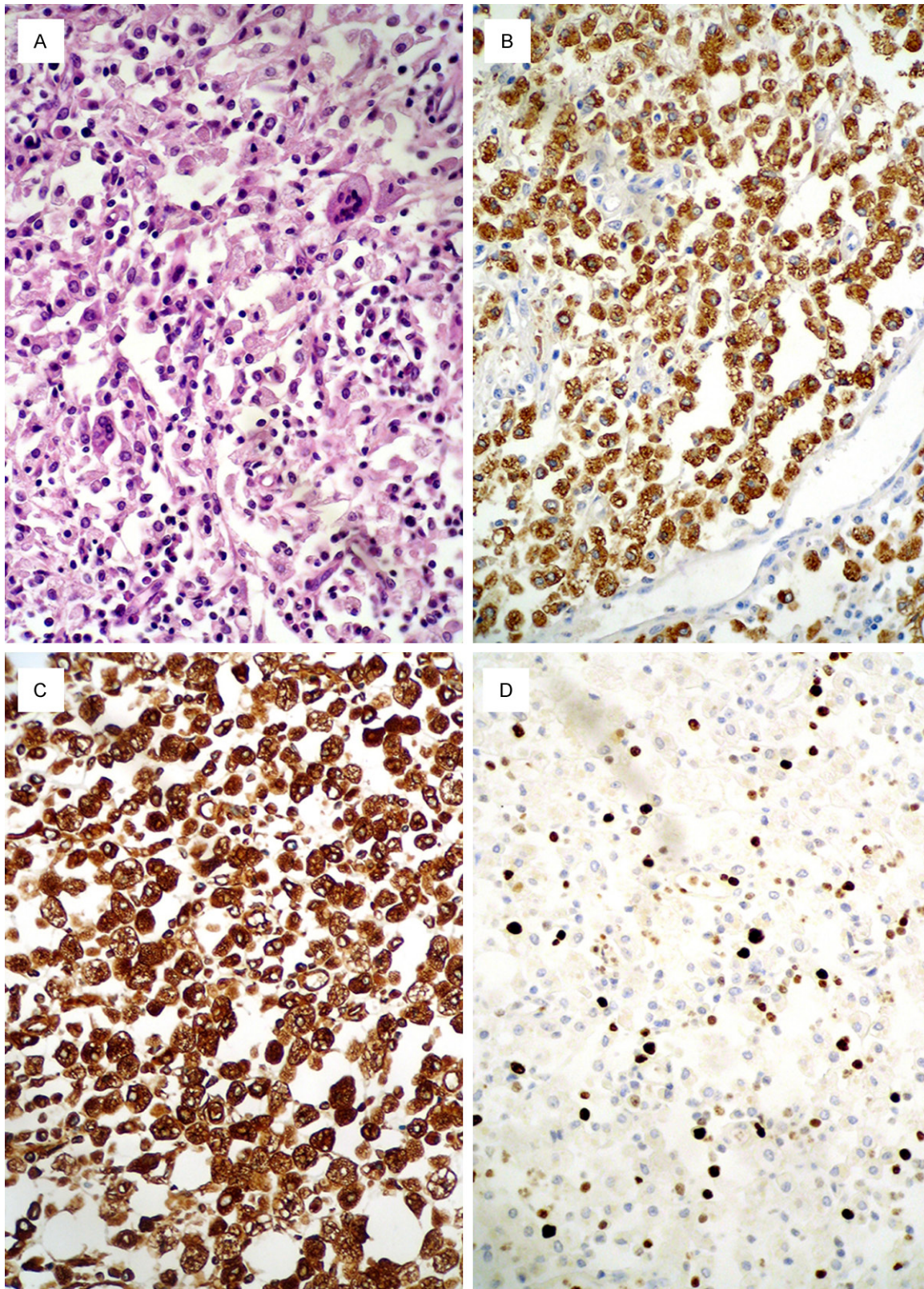


Figure 2. A. Histological findings showed XC was characterized by plenty of foamy histiocytes, eosinophils and lymphocytes infiltration (HE $\times$ 200). B. CD68 was positive to foamy histiocytes. C. Vimentin was also positive. D. Ki-67 showed a lower proliferation index ($< 10\%$). Immunohistochemistry $\times$ 200.

tion. When disease is combined with urachal remnant or adenoma, this dual pathology should be sought during surgical exploration and partial cystectomy is preferred. Therefore, doctors should take special care in diagnosing XC.

In summary, the histological features of our case were similar to those of the reported XC cases. The treatment of this case is remedied by simple excision of the lesion and a close follow-up.

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Xiang-Chun Han, Department of Pathology, The First Affiliated Hospital to Hebei North University, 12 Changqing Road, Zhangjiakou 075000, Hebei Province, P. R. China. Tel: +86-313-8043571; Fax: +86-313-8043571; E-mail: hanxiangchun1976@163.com; Dr. Ye Wang, Department of Urinary Surgery, Tangshan Gongren Hospital, 27 Culture Road, Tangshan 063000, Hebei Province, P. R. China. Tel: +86-315-3722189; Fax: +86-315-3722189; E-mail: wanyegryy@163.com

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