Xanthogranulomatous cystitis presenting as hematuria: A case report and review of literature

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Received - 12 June 2018 Initial Review - 04 July 2018 Accepted - 16 October 2018

ABSTRACT

Xanthogranulomatous cystitis (XC) is a rare benign chronic granulomatous inflammation of the urinary bladder of unknown etiology. Here, we report a rare case of XC in a 54-year-old man who presented with painless hematuria and frequent urination for the past 3 years. His urine culture and sensitivity were positive for *Escherichia coli* with significant colony count. Contrast-enhanced computed tomography showed diffusely thickened urinary bladder with no definitive mass. The patient was subjected to other investigations including cystoscopy and biopsy. All the above investigations confirmed the entity to be XC. An endoscopic resection was performed and he was started on a sulfamethoxazole-trimethoprim combination. The patient was asymptomatic at 3-month follow-up.

Keywords: *Bladder, Hematuria, Inflammation, Xanthogranulomatous Cystitis*

Xanthogranulomatous cystitis (XC) is a rare and chronic inflammatory disease of the urinary bladder with 23 cases reported so far [1]. The first case of XC was reported in 1932 by Wassiljew [2]. Histologically, xanthogranulomatous changes have been reported in other parts of the body such as the colon, ovary, pancreas, salivary gland, appendix, gallbladder, endometrium, brain, and kidney [3-5]. However, isolated urinary bladder involvement is extremely rare. We present the case of an isolated XC presenting as painless hematuria and review the literature.

CASE REPORT

A 54-year-old male, non-smoker, presented to the department with complaints of frequent urination and painless hematuria with clots on and off for the past 3 years. He had no significant medical history and his family history was non-contributory. His general examination was unremarkable and the patient's vital was stable (pulse 88, blood pressure of 120/70 mmHg, and respiratory rate 16 cycles/min). Rectal examination revealed Grade-1 benign and non-tender prostate.

Laboratory, biochemical (serum creatinine, electrolytes, and blood sugar fasting), and hematological (hemoglobin, total and differential leukocyte counts, coagulation profile, and viral markers) investigations were normal. Routine examination of the urine revealed microscopic pyuria and hematuria. His urine culture and sensitivity were positive for *Escherichia coli* with significant colony count. The patient was treated with sensitive antibiotics for 2 weeks, and then, a culture was repeated after 3 days of the cool-off period. As the pyuria and microscopic hematuria persisted despite a negative culture, three samples of urine cytology were evaluated, which was negative for malignant cells. Ultrasound abdomen was suggestive of thickened bladder wall but was otherwise unremarkable with no post-void residual urine. Contrast-enhanced computed tomography showed diffusely thickened urinary bladder with no definitive mass; no other abnormalities were detected (Fig. 1).

A provisional diagnosis of non-specific cystitis was made. Since the patient had been suffering for long despite adequate medication, he was subjected to other investigations including cystoscopy and biopsy. Cystoscopy revealed edematous, inflamed lesion on the right lateral wall of the bladder. Transurethral resection (TUR) biopsy of the lesion was performed and histopathology revealed hyperplastic transitional epithelium lining with solid nets of epithelium with occasional multinucleated giant cells and focal lymphocytic infiltration. Lamina propria showed sheets of macrophages with an eosinophilic material in the cytoplasm without Michaelis–Gutmann bodies, which was suggestive of XC (Figs. 2a and b and 3).

An endoscopic resection (partial cystectomy) was performed. Postoperatively, he was started on a sulfamethoxazole-trimethoprim combination. The patient was asymptomatic at 3-month follow-up. Repeat cystoscopy did not reveal any edema.

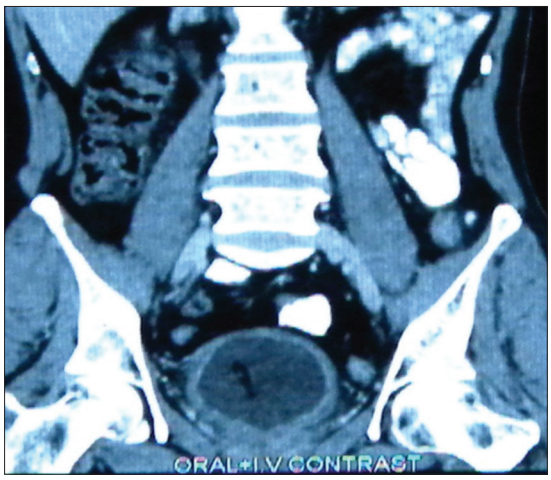

Figure 1: Contrast-enhanced computed tomography abdomen and pelvis showing diffusely thickened bladder wall

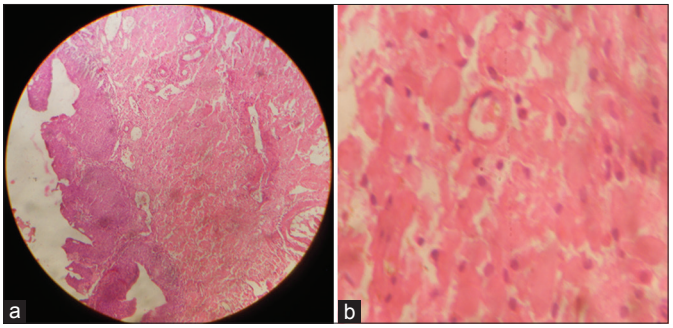

Figure 2: (a) Low-power histopathology showing transitional epithelium with sheets of histiocytes in lamina propria; (b) high-power depiction of Figure 2a

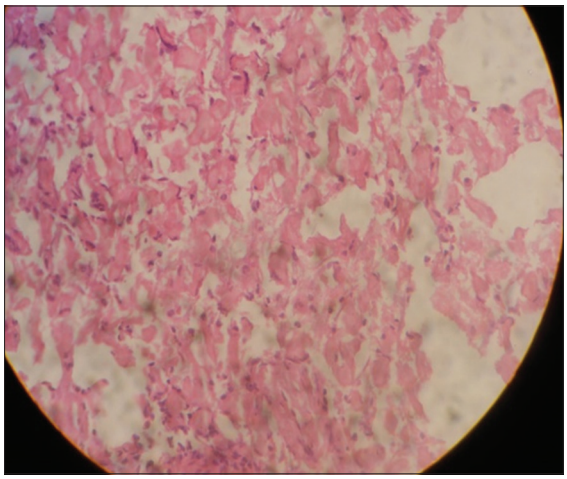

Figure 3: Picture showing sheets of foamy histiocytes with abundant cytoplasm in lamina propria

DISCUSSION

XC is a rare, benign, chronic granulomatous inflammation of the urinary bladder of unknown etiology. Various theories have been put forth, starting from abnormal lipid metabolism [6], metaplasia of urothelium due to chronic infection/inflammation [7], and immunological disorders; [8] however, the definitive cause still eludes.

The most common site of the lesion is at or near the dome of the bladder [3]. In the present case, the lesion was on the right lateral wall of the bladder which is rare. XC may exist alone or with bladder malignancy [5]. Clinically, it does not have any specific presentation. It may present as persistent lower abdominal pain and cystitis-like symptoms, umbilical discharge, and occasional hematuria, thus mimicking a tumor of the urinary bladder [9]. Rarely, it may be associated with inflammatory bowel disease [10]. Majority of the cases reported in the literature have been associated with urachal diverticulum [11]. The optimal treatment for XC is not known; however, surgical resection (partial cystectomy) is preferred [10]. We subjected our patient to endoscopic resection followed by broad-spectrum antibiotic therapy. At 3-month follow-up, the patient was fine.

Histologically, XC need to be differentiated from malakoplakia which is characterized by Michaelis–Gutmann bodies, whereas in the case of XC, there are lipid-laden macrophages, plasma cells, and lymphocytes [10].

CONCLUSION

XC is a rare cause for painless hematuria which can be safely managed with endoscopic resection and suppressive post-operative antibiotic therapy.

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Funding: Nil; Conflict of Interest: Not Declared.

How to cite this article: Gupta R, Arti, Kapoor B. Xanthogranulomatous cystitis presenting as hematuria: A case report and review of literature. Indian J Case Reports. 2018;4(5):403-404.

Doi: 10.32677/IJCR.2018.v04.i05.024