Case Report

Extensive xanthogranulomatous cystitis mimicking bladder cancer

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Abstract Xanthogranulomatous cystitis (XC) is a rare benign disease of unknown etiology. A 39-year-old female presented with 2 month history of urgency, dysuria, lower abdominal mass. On physical examination a hard hypogastric mass was present fixed to the rectus muscle. Computerized tomography (CT) abdomen showed heterogeneous enhancing mass arising from the anterior bladder wall with infiltration of the overlying parietal wall. Cystoscopy revealed extensive growth involving the entire wall of the bladder. A biopsy showed cystitis with focal areas suggestive of urothelial neoplasia of unknown malignant potential. Suspecting bladder cancer, we proceeded with radical cystectomy with ileal conduit. Histopathology revealed cystitis cystica with XC of the entire bladder. This is, to our knowledge, the first time that a case has been found to be so extensive with infiltration of the parietal wall and second time that radical cystectomy has been performed for XC.

Key Words: Partial cystectomy, radical cystectomy, urachal remnant, xanthogranulomatous cystitis

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INTRODUCTION

Xanthogranulomatous change is characterized histologically by the presence of xanthoma cells (lipid-laden macrophages), multinucleated giant cells and cholesterol clefts. This change has been reported to occur in many sites, including the colon, ovary, pancreas, salivary gland, appendix, gall bladder, endometrium, brain and kidney.[1] However it is rarely reported in the bladder. We report one such case and discuss the treatments available.

CASE REPORT

A 39-year-old female presented with two months history of urgency, dysuria, lower abdominal pain and lower abdominal swelling. There was no history of hematuria. On physical examination a hard hypogastric mass was present fixed to the rectus muscle. It was bimanually palpable. Routine blood investigations were normal. Urine routine was normal and culture was sterile. Ultrasonography showed a heterogeneous mass lesion from the anterior wall of the bladder. Contrast-enhanced computerized tomography (CECT) abdomen showed a heterogeneous enhancing mass 8.8 by 4.4 cm mass arising from the anterior bladder wall and dome, extending to the umbilicus with infiltration of the overlying parietal wall [Figure 1]. Cystoscopy revealed extensive growth involving the entire anterior wall and dome of the bladder extending to posterior wall. A biopsy done revealed cystitis with focal areas suggestive of urothelial neoplasia of unknown malignant potential. Metastatic workup was normal. As we could not rule out the presence of bladder tumor and due to the large size on the lesion, we proceeded with radical cystectomy with ileal conduit. The mass was found to be arising from the entire wall of the bladder and was infiltrating into the posterior rectus sheath and muscle, which needed to be partly resected along

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with the specimen [Figure 2]. Histopathology revealed cystitis cystica with xanthogranulomatous cystitis (XC) of the entire bladder wall [Figures 3 and 4]. There was no malignancy or urachal remnant. The post-operative course was uneventful and at 1 year follow up the patient is doing well.

DISCUSSION

XC is a rare benign inflammatory disease and majority of the reported cases are associated with urachal remnant or adenoma.[1] Twenty-seven cases have been reported in the literature, with the following clinical characteristics: Median age of 42 years (range, 16 to 76 years); no sexual predilection (13 cases have been of females); and the majority of lesions were located in the bladder dome (18 cases). Most cases are associated with aurachal remnant (17 cases). In the kidney xanthomatous change almost always develops in response to chronic low grade inflammation, often as a response to obstruction of urine.[1] The etiology of XC is unclear. Proposed explanations suggest a chronic inflammatory process caused by mechanisms such as (i) Immunological defect of the macrophage,[2] (ii) chronic infection of the urachal diverticulum or cyst,[3] (iii) gram negative or anaerobic bacteria such as in urinary tract infections,[4] or infection after tubal ligation,[5] (iv) foreign material such as retained suture material,[4] (v) local response to a bladder tumor,[2] and (vi) abnormal lipid metabolism and lipid accumulation in a macrophage. Histologically xanthogranulomatous lesions can be confused with malakoplakia and can be differentiated from it, by the absence of Michaelis-Guttmann bodies (basophilic lamellar inclusion bodies) and presence of large no of monocytes.[4] The symptoms of XC are nonspecific and difficult to distinguish from other disorders of the bladder, especially malignancy. XC most often presents with lower abdominal mass and symptoms of cystitis, such as frequency,

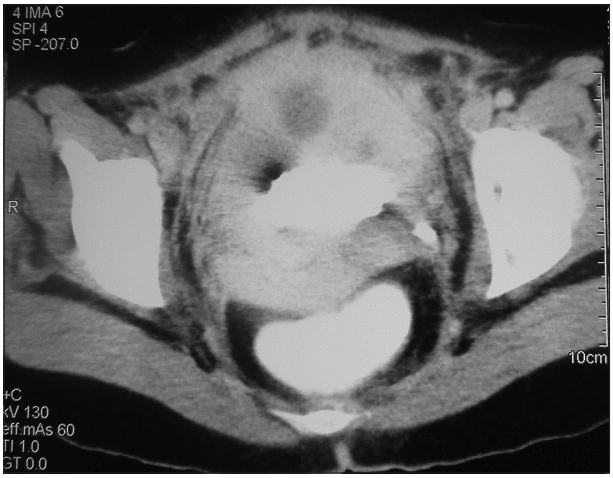

Figure 1: Axial CECT section of the pelvis showing heterogeneously enhancing diffuse wall thickening of the bladder with infiltration of the parietal wall. The arrow shows a well-defined enhancing cystic area in the anterior wall of the bladder

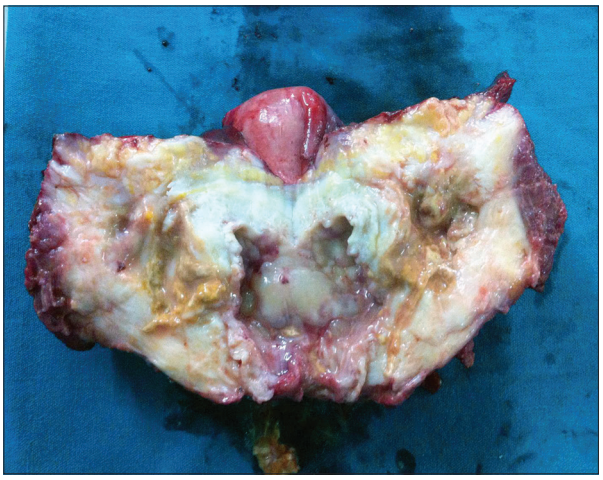

Figure 2: Gross findings revealed thickening of the entire bladder wall extending anteriorly into the posterior rectus sheath. A cystic cavity was seen in the anterior wall of the bladder

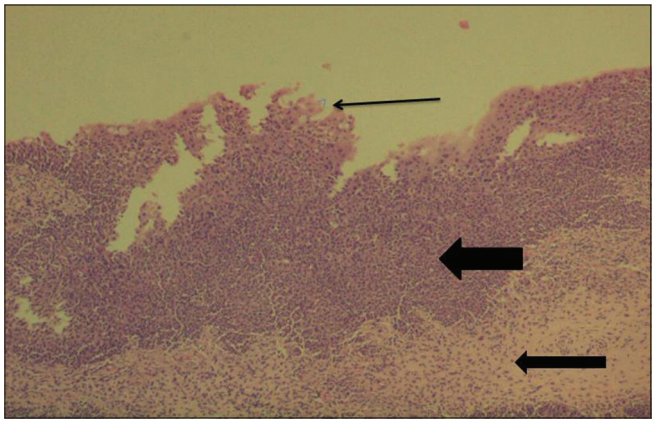

Figure 3: Hematoxylin and eosin section; x10, thin arrow - squamous metaplasia, medium-size arrow-sub-epithelial area with foamy histiocytes, thick arrow-transitional epithelium with inflammatory infiltrate

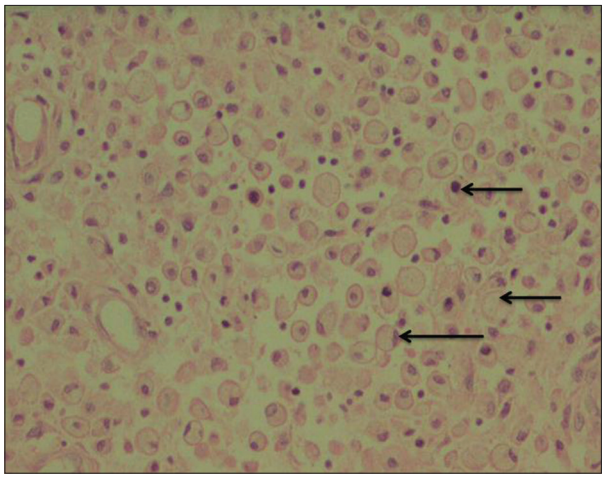

Figure 4: Hematoxylin and Eosin; x40, the arrows show foamy histiocytes with abundant clear feathery cytoplasm with nuclei

urgency, and dysuria.[6] XC may be associated with carcinoma and can also mimic it.[2] Hence, it is important that the surgeon should be aware of the potential dual pathology and tumor should always be sought if this unusual condition is diagnosed in the bladder. Also, it is essential the pathologists should sample of the entire surgical specimen during gross examination. Although a trial of long-term broad-spectrum antibiotics has been suggested, medical treatment has not been found to be effective. As most of the patients had an associated urachal remnant, and as it is difficult to differentiate it from cancer, conservative treatment is rarely employed. The curative treatment of choice is surgery.[7] Localized disease or that involving the dome of the bladder may be amenable to simple excision of the lesion. However, when the disease is combined with urachal remnant or adenoma, partial cystectomy is preferred and is the most commonly performed procedure. Endoscopic excision has also been reported for small lesions.[8,9] To our knowledge there has been only one report of radical cystectomy being performed.[10] Our case was not suited for partial cystectomy or endoscopic resection due to large size with extension to the parietal wall, and as we could not rule out malignancy, we proceeded with radical cystectomy.

CONCLUSION

When a patient presents with a large bladder mass with significant symptoms and biopsy is not conclusive for malignancy, one should suspect chronic inflammatory pathology of the bladder. In such a situation radical cystectomy is a reasonable option due to high probability of bladder malignancy and lack of any beneficial effect with medical management even if it is a benign inflammatory pathology.

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