Extensive xanthogranulomatous cystitis mimicking bladder cancer

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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. 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. 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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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.

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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 urachal 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,
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