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Xanthogranulomatous urethritis and cystitis: a rare clinical and pathological entity

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ABSTRACT

Xanthogranulomatous inflammation of the urethra and the urinary bladder is a very rare pathological condition characterized by a chronic inflammatory infiltration composed mainly of foamy macrophages, with the presence of multinucleated giant cells. In a clinical examination, it can mimic urinary bladder carcinoma. This report presents the extremely rare case of a co-existing xanthogranulomatous urethritis and cystitis in a 64-year-old woman with recurrent dysuria, and with the suspicion of malignancy – as indicated on the basis of a cystoscopic examination. The standard treatment of this disorder is surgical resection, but in the presented case, only a diagnostic biopsy was performed. Because of the persistence of clinical symptoms, a cystoscopic examination and biopsy was repeated three times in a two year period. There was no malignancy seen in the repeated biopsies.

INTRODUCTION

Xanthogranulomatous inflammation of the urethra and the urinary bladder is a very rare pathological condition characterized by a chronic inflammatory infiltration composed mainly of foamy macrophages, with the presence of multinucleated giant cells. Clinically, it may be mistaken for carcinoma or another malignancy involving the urethra [1]. Until recently, less than 50 cases of this type of inflammation in the urinary bladder have been described [8], and, based on PubMed, only one case of xanthogranulomatous urethritis was reported. In this work, a new extremely rare case of concomitant xanthogranulomatous urethritis and cystitis is presented.

CASE REPORT

A 64-year-old woman came to us with a two year history of recurrent dysuria, i.e. increased frequency and urgency of urination, decreasing stream of urine and a feeling of incomplete emptying of her bladder. Physical and urine examination, as well as a urodynamic procedure was performed. The patient was referred to our hospital with the suspicion of urethral stenosis. A cystoscopic examination revealed a diffuse exophytic growth located in the urinary bladder cervix and in the proximal part of the urethra. However, the dome of the bladder and the vesico-ureteric junction were uninvolved. With the suspicion of malignancy, a biopsy was taken from the posterior wall, the cervix of the urinary bladder and from the urethra.

A microscopic examination showed a diffuse infiltration composed of foamy macropages, multinucleated giant cells and lymphocytes, with the formation of granulomas accompanied by fibrosis (Fig. 1A). The presence of macrophages was confirmed by immunohistochemical reaction with CD68 (Dako) (Fig. 1B). Additional histochemical stainings (PAS, Grocott, Warthin-Starry) were used in differential diagnosis.

The postoperative course was uneventful, and the patient was treated by way of antibiotic administration. Because of persistent clinical symptoms, the biopsy was repeated three times in a two year period. With uroepithelial carcinoma being suspected, the second biopsy was performed 10 months after the first, while half a year later, the biopsy was repeated because of uretheral stenosis. The last cystoscopic examination was made after 10 months, with clinical diagnosis of an inflammatory tumor. The histological picture of two middle examinations showed chronic inflammatory infiltration with unspecific granulation. The last examination revealed features similar to the first specimen, additionally displaying dense lymphocytic and plasmocytic infiltration and single multinucleated giant cells. There was no malignancy in any of the biopsies.

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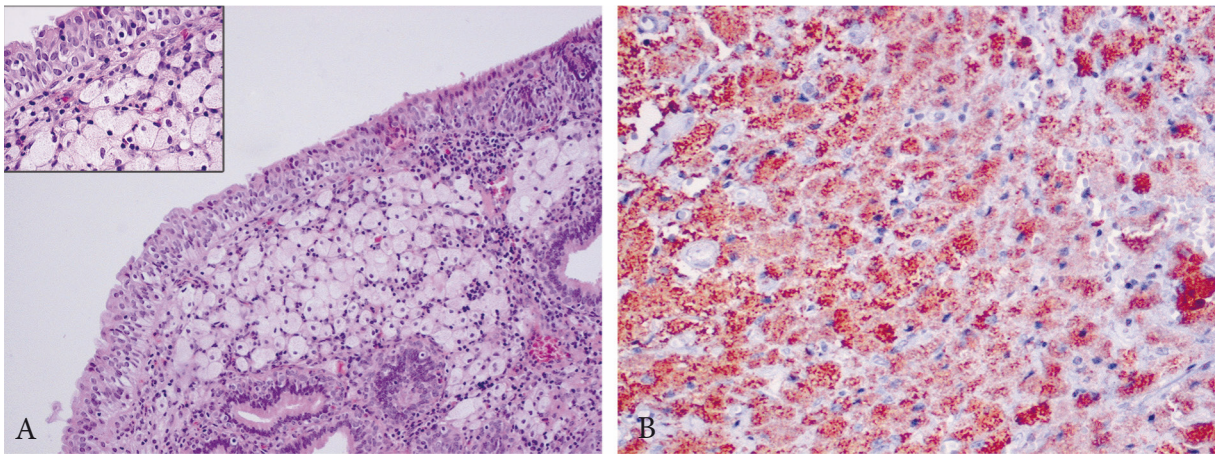

Figure 1. Xanthogranulomatous urethritis. A. The prominent collection of foamy histiocytes in the lamina propria immediately subjacent to the uroepithelium (H&E, objective magn. 20×; islet 40×). B. Positive immunostaining for CD68 in foamy macrophages (objective magn. 20×)

DISCUSSION

Xanthogranulomatous inflammation is a common lesion observed in many organs – especially the gallbladder, pancreas, appendix, colon, ovary, endometrium, brain and kidney [11]. A location in the urinary bladder is uncommon, but in the urethra, this is exceptional [10].

Although the first case of xanthogranulomatous cystitis was described in 1932 [14], the etiology of this disorder is unclear. It may be associated with the chronic inflammation of the urachal diverticulum [9], with a cyst [3] or with adenoma [6]. Predisposing factors are also a chronic infection induced by the Gram-negative or anaerobic bacteria typical for urinary tract infections [5] or an infection that comes about after tubal ligation [13]. Other causes could be related to a defect in macrophages function [2], abnormal lipid metabolism or an accumulation of lipids in macrophages [12]. An association with inflammatory bowel disease has also been described [3]. Xanthogranulomatous cystitis can be regarded as a type of response for inert foreign materials such as retained suture [4,5] or as a local response to a bladder tumor [2].

Symptoms of xanthogranulomatous cystitis and urethritis are nonspecific and difficult to be distinguished from other diseases of the urinary bladder and urethra. Moreover, the gross appearance (irregular, exophytic mass) may simulate a carcinoma (which is much more common lesion in this location) [2]. Pathological examination is, hence, essential for diagnosis. The microscopic features of xanthogranulomatous cystitis and urethritis are very characteristic, but require careful differential diagnosis. This is especially true with regard to malakoplakia, as this can produce similar clinical settings. In the reported case a diagnosis of malakoplakia was excluded by negative periodic acid Schiff (PAS) reaction and lack of characteristic Michaelis-Guttman bodies. The differential diagnosis should also cover other causes of this type of inflammatory reaction. In the reported case, bacterial and fungal infection was excluded by histochemical stainings (Grocott, Warthin-Starry). Surgical resection is a curative treatment for xantogranulomatous inflammation [7], but in the presented case, a diagnostic biopsy alone was performed. Xanthogranulomatous cystitis usually involves the dome of the bladder, and in such cases, may be amenable to simple excision. Small lesions are removed during endoscopy [1]. In the case of the involvement of the bladder cervix and the urethra, complete surgical resection is impossible due to technical reasons.

Because xanthogranulomatous cystitis and urethritis may imitate bladder cancer, and the coexistence of xathogranulomatous inflammation with carcinoma has been reported [2], this dual pathology should be sought out during surgical exploration and histopathological assessment.

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