

A Case of Genitourinary Xanthogranulomatous Inflammation

Sheng Tai and Chaozhao Liang*

The Department of Urology, the First Affiliated Hospital of Anhui Medical University, the Urological Institute of Anhui Medical University, China

Abstract

A case of xanthogranulomatous salpingitis associated with a little invasive lesion of cyst in a 52-years-old woman is presented. Xanthogranulomatous inflammation is a rare form of chronic inflammation, destructive to the involved organs. It is characteristic of the presence of plasma cells, neutrophils, lymphocytes, lipid-filled macrophages. It is reported that most patients with xanthogranulomatous salpingitis often have a clinical history of pelvic inflammatory disease, endometriosis, etc. Here, we reported a case is with unilateral xanthogranulomatous salpingitis with invasion of cyst.

Keywords: Xanthogranulomatous; Salpingitis; Cystitis

Introduction

The xanthogranulomatous inflammation is an extremely rare inflammatory lesion, characteristic of destructive to the normal tissue of the affected organs, the presence of a large number of lipid-containing macrophages with an admixture of lymphocytes, plasma cells, and neutrophils, presence of multinuclear giant cells, etc [1]. It is debilitating that only a few reports have been reported in the Chinese medical literatures. Multiple organs have been documented to be affected by xanthogranulomatous inflammation, most commonly involving kidney followed by cholecyst [1]. Involvement of genitourinary tract is also rare. The mechanism of this form of inflammatory entity is poorly understood. Pelvic Inflammatory Disease (PID), especially the genital tract infection, is one of the most attributable culprit [2]. Other more general contributory factors in the pathogenesis of xanthogranulomatous inflammation may include necrosis, hemorrhage, and obstruction [3]. Here we reported a fortuitous case individual suffering from unilateral xanthogranulomatous salpingitis with infiltration of cyst, inducing the xanthogranulomatous cystitis.

Case Presentation

A 52-year-old woman, with no past notable medical history, present with urgency, frequency, lower abdominal mass and pain, constipation for 1 month's duration. She had been administered with antibiotic for acute lower urinary tract infection for 2 weeks, but there is not any improvement. She was gravid 2, para 1 and her LMP was 3 weeks prior. On physical examination, she was lethargy and afebrile. The bimanual pelvic examination revealed a right suprapubic mass (7×6 cm) was palpable, characterized by rounded, smooth, firm, non-tender.

Routine blood examinations, including normal hematological and biochemical markers, were all performed. Testes showed the presence of inflammation with slightly increased total white blood cells ($13.28 \times 10^9/L$) and an elevation of C-reactive protein (78 mg/L). Tests for carbohydrate antigen 125 (CA125), carbohydrate antigen 199 (CA199), carcinoembryonic antigen (CEA), and Alpha Fetoprotein (AFP) were all negative. Urinalysis demonstrated 1-2 white blood cells per high power field and no malignant cells. Urine culture was negative for bacterial growth. The vaginal *Escherichia coli* culture is positive. The trans-abdominal ultrasounds revealed a heterogenic cystic tubular structures, approximate 8.5 cm×7.4 cm on the right annex. The enhanced CT scan confirmed this lesion and revealed a mass with fluid at right cyst (8.5×8 cm) (Figure 1).

Under the general anesthesia, the patient was offered the diagnostic laparoscopy for the annex mass lesion. Because pathological examination revealed the inflammation lesion for annex lesions, the right salpingo-

ophoretomy was performed with pathologic confirmation. Meanwhile, the partial laparoscopic cystectomy was also performed. Examination of histological pathology revealed a couple of specimens of abundant eosinophils, plasma cell, lymphocytes, neutrophils, multinuclear giant cells (Figures 2A, 2B). Many of the histocytes in the lamina appeared to be containing abundant lipid-like material. All these features are consistent with the characteristic of xanthogranulomatous salpingitis and cystitis [4-7].

Discussion

The xanthogranulomatous lesion is a rare chronic inflammatory disease characterized by the focal or diffuse destructive inflammatory process, with accumulation of lipid-laden fibrous tissue, and acute and chronic inflammatory cells; It may involve any organ, but the most common sites are known as the gallbladder and kidney [8]. The xanthogranulomatous inflammation involving the salpingitis and cystitis simultaneously is very rare. However, it may be



Figure 1: Enhanced computed tomography scan of abdomen and pelvis reveals that an 8.5×8 cm mass (arrows) in the right pelvic with scatter rim enhancement in the right annex. The lesions invade the right side of cyst.

*Corresponding author: Chaozhao Liang, The Department of Urology, the First Affiliated Hospital of Anhui Medical University, the Urological Institute of Anhui Medical University, 218 Jixi, Ave., Hefei, Anhui, 230022, China, E-mail: taishengwk@163.com

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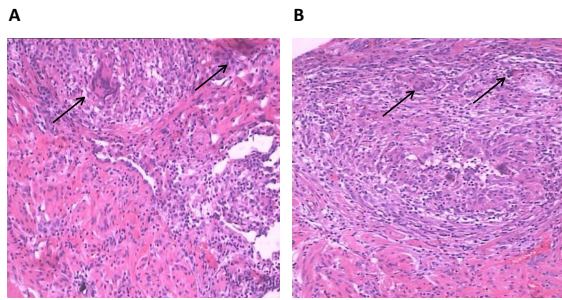


Figure 2: Hematoxylin-eosin staining of annex (A) and cyst (B) lesions shows an increase of plasma cells and eosinophils, and xanthoma cells (arrows) with granulating inflammations (H&E 100 X (A,B)).

difficult to differentiate from infiltrative cancer because features of xanthogranulomatous inflammation are represented in the form of an irregular mass-like lesion with fibrosis and inflammation extending the surrounding tissue [9]. Therefore, the appropriate surgical approach for pathological diagnosis is needed.

The xanthogranulomatous lesions are unusual forms of chronic inflammatory process, pathologically characterized with presence of large lipid macrophages, multinuclear gigantic cells, etc [5,6,10]. The exact pathogenesis of xanthogranulomatous inflammation remains to be debated. Many unrelated disorders may have the same mechanism of foam cell proliferation, infection, poorly antibiotic therapy, abnormality of lipid metabolism, endometriosis, etc. Such particular lesions often involved in a wide spectrum sites, the mandible, retroperitoneum, lateral ventricle, orbit, hepatic, stomach, kidney, ovary, etc [11]. Some reviews of the literature revealed a couple of cases of fallopian tube involvement reported to date [11]. But the association of this entity with xanthogranulomatous cystitis is interesting in a manner that it is extremely rare yet does not develop in a large number of cases. The xanthogranulomatous cystitis may present with urinary symptoms, abdominal pain or umbilical discharge [12]. This condition is characterized microscopically by multinucleated giant cells, lipid-laden macrophages (xanthoma cells) and cholesterol crystals [12]. Macroscopically, it manifests as soft yellow-brown plaque, and malakoplakia is an inflammatory condition attributed to abnormal macrophage response to *Escherichia coli* urinary infection and has a similar clinical picture and histological change. Malakoplakia may be characterized by basophilic lamellar inclusion bodies (Michaelis Gutmann bodies) and large aggregates of monocytes (Von Hanseman bodies) [12]. However, this case is negative for urine test and culture. The reason why the urine test and culture is negative is that xanthogranulomatous cystitis is secondary to the unilateral xanthogranulomatous salpingitis.

The exact pathogenesis of xanthogranulomatous inflammation remains to be controversial. Many unrelated disorders may have the same mechanism of foam cell production. Pelvic inflammatory disease, endometriosis, poor antibiotic therapy and radiotherapy are the predisposing conditions for the development of xanthogranulomatous salpingitis [2,4]. Proposed explanations suggest a chronic inflammatory process caused by mechanisms such as: immunological defect of the macrophage [13]; chronic infection of the urachal diverticulum or cyst [14]; gram negative or anerobic bacteria such as in genitourinary tract infections or infection after tubal ligation [14]; foreign material such as retained suture material [10]; local response to a bladder tumor and abnormal lipid metabolism and lipid accumulation in a macrophage [15-17]. Consistent with other studies, this patient's vaginal *Escherichia coli* culture is positive, and previous tubal ligation

with local inflammation could be a predisposing cause. Co-exist factors may also be recognized in some cases. The bleeding and obstruction may predispose to this infection, tissue necrosis occurrence, and then followed by the release of cholesterol and other lipids and phagocytosis by macrophages. On the other hand, in some of the previous literatures, the xanthogranulomatous inflammation has been linked to the intrauterine contraceptive devices [18]. Consistent with other reports, in this present case, the patient did have her IUD for 6 years duration, which may contribute the initiation or exacerbation of xanthogranulomatous lesion.

In the present case, we demonstrated that our patient presented with unilateral xanthogranulomatous salpingitis with invasion of cyst. More importantly, the pathologic examination of invaded cyst revealed the xanthogranulomatous lesion characteristics (Figure 2B). The patient did not have any history of a lipid metabolic disease, nor did her history give an indication of immunodeficiency.

This case report showed the xanthogranulomatous inflammation can involve the fallopian tube and bladder simultaneously. This patient presented with urinary tract infection symptoms and the xanthogranulomatous lesion can also be attributable to the infection [19,20]. Xanthogranulomatous salpingitis represents a chronic form of inflammation secondary to a wide spectrum of factors and should be considered to be the differential diagnosis of unilateral or bilateral pelvic masses. On the other hand, the lower urinary tract symptoms (urgency, frequency, odynuria) are often present with among urologic outpatients and the xanthogranulomatous cystitis is also considered to be one of the differential diagnoses in the future.

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