# *Xanthogranulomatous cystitis: case report and literature review*

**AUGUSTO RAMOS PRADO** – Médico Especialista em Urologia. Professor Titular da Disciplina de Urologia da Universidade Federal de Santa Maria.
**RAFAEL CRISTIANO GEISS SANTOS** – Acadêmico de Medicina da Universidade Federal de Santa Maria. Doutorando de Medicina da Universidade Federal de Santa Maria.
**LUANA PIZARRO MENEGHELLO** – Acadêmica de Medicina da Universidade Federal de Santa Maria. Doutoranda de Medicina da Universidade Federal de Santa Maria.
**CAMILA HUBNER DALMORA** – Acadêmica de Medicina da Universidade Federal de Santa Maria. Acadêmica de Medicina da Universidade Federal de Santa Maria.

Universidade Federal de Santa Maria (UFSM).

✉ Endereço para correspondência:
**Rafael Santos**
Av. Presidente Vargas 475
97020-001 – Santa Maria – RS, Brasil
☎ (55) 9131-1070
💻 uro.husm@gmail.com

**RESUMO**

A cistite xantogranulomatosa (CX) é uma rara doença inflamatória crônica e benigna de etiologia desconhecida. É apresentado um caso de cistite xantogranulomatosa em um paciente masculino de 53 anos. A evolução do paciente incluiu estudos clínicos, radiológicos e histológicos. Não houve recorrência em 32 meses de seguimento após o tratamento. Este caso é provavelmente o 21º a ser relatado no mundo, e o primeiro caso na literatura brasileira.
Palavras-chave: Cistite Xantogranulomatosa, Doença Inflamatória, Etiologia Desconhecida.

UNITERMOS: Apendicite Aguda, Ultra-Sonografia, Abdômen Agudo Inflamatório.

**ABSTRACT**

*Xanthogranulomatous cystitis (XC) is a rare benign chronic inflammatory disease of unknown etiology. A case of Xanthogranulomatous cystitis in a 53 year-old male is presented. Patient evolution included clinical, radiological, and histological studies. He had no recurrence of XC 32 months after treatment. The present case is probably the 21st to be reported in the world, and the first case in the Brazilian literature.*

KEYWORDS: *Xanthogranulomatous Cystitis, Inflammatory Disease, Unknown Etiology.*

## INTRODUCTION

Xanthogranulomatous change has been reported to occur in many sites (1, 2), including the colon, ovary, pancreas, salivary gland, appendix, gallbladder, endometrium, brain, and kidney (3). However, xanthogranulomatous cystitis is a rare, benign chronic inflammatory disease of unclear etiology and was first described in 1932 (4). Later on only single cases or small series of cases of this disease were reported, most of which were described in the Japanese literature (3, 5, 6). We describe herein the first case in Brazilian literature.

## CASE REPORT

A 53-year-old man presented in March 2004 with a one-month history of urgency, frequency, micturation pain, and lower abdominal pain. He had no significant past medical history. Physical examination revealed a rounded, smooth mass in the suprapubic area. The first diagnostic hypothesis was urinary retention, but the urethral catheterization failed in making the mass disappear. Then, solid tumor was the most probable diagnosis. Laboratory studies included normal hematological and biochemical profiles, except for C-reactive protein elevation in the peripheral blood (3.6 mg/dL; reference < 0.5 mg/dL). Tests for carcinoembryonic antigen (CEA), carbohydrate antigen 125 (CA 125) and carbohydrate 19.9 (CA 19.9) were all negative. Urinalysis showed 8-17 red and 10-20 white blood cells per high power field. Urine culture was negative, and urine cytology revealed no malignant cells. Excretory urography showed a normal upper urinary tract. Cystoscopy showed a normal urothelium. Computed tomography (CT) demonstrated diffuse thickening of the bladder walls, but no mass evidence (Figure 1). Due to the potential of malignancy, laparotomy was elected. It confirmed the diffuse thickening of bladder walls, and revealed a perivesical mass. Biopsy of the mass and random biopsies of the walls was realized. No urachal remnant was found. Histological examination of the specimen (Figure 2) revealed chronic inflammatory xanthogranulomatous cystitis, with no evidence of malignant cells. Postoperative course was uneventful. The patient received antibiotics for 7 days, was discharged home using SMZ+TMP for 2 months, and remains asymptomatic with no recurrence of XC after 32 months of follow up.

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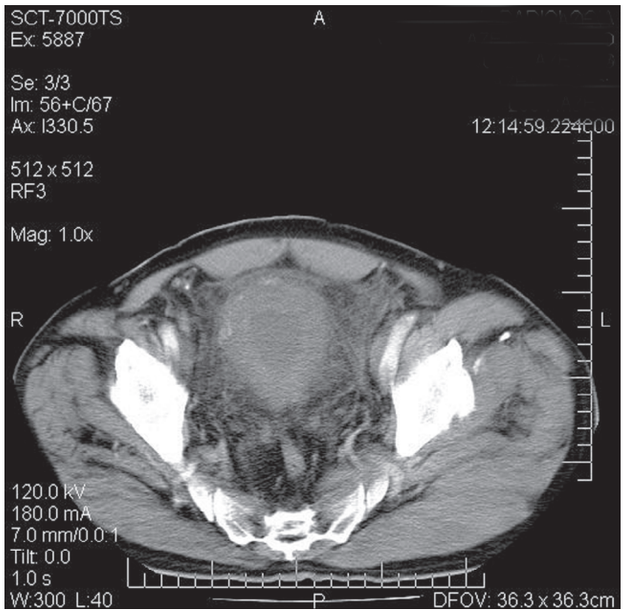



**Figure 1** – Computed Tomography of the pelvis shows diffuse thickening of the bladder walls.

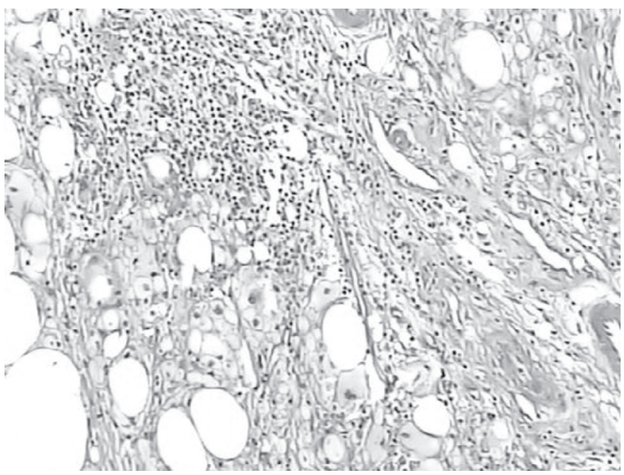


**Figure 2** – Chronic xanthogranulomatous inflammation, with increase of plasma cells, eosinophils, and xanthoma cells. (H&E X 400).

## DISCUSSION

Xanthogranulomatous cystitis is a rare benign inflammatory disease, and majority of the reported cases are associated with urachal diverticula (7). It was first described in the medical literature in 1932 by Wassiljew (4). Since then, 20 cases have been reported worldwide, the majority of cases in the Japanese literature (1, 4-6, 8-10).

The etiology of XC is unknown. However, there are many theories as to its origin, such as immunological disorders (5, 10); abnormal lipid metabolism (11); metaplasia of the urothelium due to chronic infection (12). Other possible etiologies involve an abnormal host response to tumor (1) and reduction of chemotatic activities (5). The etiology that XC leads to a chronic inflammation of an urachal cyst was posed because XC contained an urachal remnant in 15 (75%) of 20 cases and 76% of XC occurred at the dome of the bladder. In some cases, XC may be associated with some chronic inflammation without an urachal cyst (3).

A review of literature revealed 20 previous cases of XC reported and confirmed by biopsy (1, 3, 4, 8-10). The following characteristics have been reported in the 21 cases, including the present case: median age, 42 years (range 16-76); males 12; females 8; one case with unknown sex and age; Japanese patients, 11 (majority of cases). The location of XC was mainly at the dome of the bladder in 14 (66%) of cases, near the dome in three and unknown in the remaining cases.

Xanthogranulomatous cystitis most often presents with urinary symptoms that are difficult to differentiate from those of other bladder problems, and most cases are associated with an urachal remnant (6, 7, 9). The main symptoms found in the review were low abdominal palpable mass, cystitis-like symptoms, umbilical pus discharge, and hematuria (1, 3, 4, 8-10).

Since medical treatment has not been found to be effective, and 80% of the patients had also urachal remnant, conservative treatment was rarely employed and generally unwise (9). The curative treatment of choice is surgical resection (5, 9). Localized disease or one involving the dome of the bladder may be amenable to simple tumor excision. However, when the disease is combined with urachal remnant or adenoma, partial cystectomy is preferred (9). Chronic suppressive antibiotic therapy and urinary astringents may be helpful. Surgery is necessary for excision of urachal lesions to rule out occult adenocarcinoma. Routine excision of isolated xanthogranulomatous lesions may not be indicated (3, 5). Sometimes it is difficult to distinguish between XC urachal adenoma and XC accompanying carcinoma before operation; therefore, doctors should take special care in diagnosing XC (1, 13).

In one report, Goel et al. (14) described a suggestive case of XC. A 50-year-old man presented with urgency, frequency, hematuria and lower abdominal pain. Cystoscopy revealed inflamed bladder with multiple small polypoidal growth that had histological findings suggestive of XC on biopsy. Patient was treated with augmentation cystoplasty and prolonged administra-



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tion of broad spectrum antibiotics. The patient remains asymptomatic at 1-year follow-up.

Microscopically, Xanthogranulomatous lesions are differentiated by multinucleated giant cells (Touton cells). Occasionally, cholesterol crystals are seen. Malacoplakia is recognized by the presence of Michaelis-Gutmann bodies. Histologically, these lesions can be confused with clear cell adenocarcinoma of the kidney. (11, 15). They also have been misdiagnosed as benign cystic adenomatous renal parenchymal tumors (5).

Xanthogranulomatous cystitis has a benign presentation, with no tendency to recidivate, according to follow-ups of more than 15 years (9). In our case the patient is asymptomatic and has been well more than 32 months after surgical intervention.

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