

Xanthogranulomatous Cystitis: A Case Report and Clinicopathological Review

Dear Editor,

Xanthogranulomatous cystitis is an unusual benign, chronic inflammatory condition. Xanthogranulomas are mostly reported in the kidney,¹ rarely presented in gynaecological literature.

Case Report

A 53-year-old woman presented with asymptomatic microscopic haematuria. Physical examination was unremarkable. She had a previous laparoscopic tubal ligation after 2 normal vaginal deliveries. Cystoscopy showed extrinsic compression of the bladder dome by a pelvic mass. An intravenous pyelogram showed a round calcification in the right hemipelvis. A computed tomography scan of the abdomen and pelvis demonstrated a heterogenous mass, 56 mm x 50 mm with a 28 mm cystic component, adherent to and indenting the antero-superior wall of the bladder with a 35 mm posterolateral uterine leiomyoma (Fig. 1). Urine cytology was negative for malignancy. Urine culture reported no bacterial growth. A pelvic ultrasound showed a left adnexal 69 mm x 54 mm complex mass compressing the left lateral bladder wall. Two intramural uterine leiomyomas, measuring 56 mm x 37 mm and 40 mm x 32 mm were also noted. A complete blood count, blood serum chemistry panel, cancer antigen 125 and carcinoembryonic antigen (CEA) were within normal limits.

The diagnostic impression was a possible parasitic uterine leiomyoma or an endometriotic lesion on the bladder. The patient underwent an exploratory laparotomy. Intraoperatively, an 8-cm firm, smooth and well circumscribed mass was noted in the space of Retzius, involving the muscularis of the bladder dome (Fig. 2). The uterus was enlarged with 2 uterine leiomyomas. Cystoscopy showed egress of urine through both ureteric orifices. A total abdominal hysterectomy, bilateral salpingo-oophorectomy with partial cystectomy was performed with bladder repair.

Histopathological diagnosis of the bladder mass was reported as xanthogranulomatous inflammation of bladder wall—described as sheets of xanthomatous necrotic elements and chronic inflammatory infiltrate with granulation tissue replacing the surface of muscular wall tissue (Fig. 3). There were no well-formed epithelioid granulomas or malakoplakia. The mass was described as



Fig. 1. Computed tomography of the pelvis showing a heterogenous mass (arrow) adherent to and indenting the anterior and superior wall of the bladder (B), with a cystic component within the mass. The uterus (U) was also noted to be enlarged with a leiomyoma.

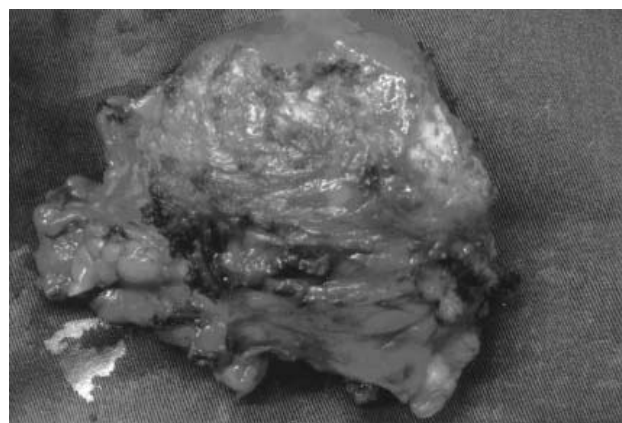


Fig. 2. Excision specimen showing a well circumscribed mass, 95 mm x 65 mm x 60 mm, arising from part of the thickened bladder wall.

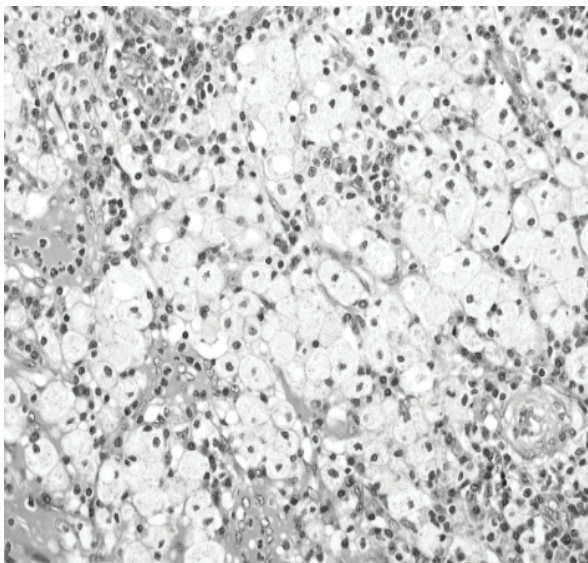


Fig. 3. The specimen is characterised by sheets of foamy cells or macrophages in a background of numerous plasma cells and lymphocytes.

a cyst filled with yellow-orange thick fluid with a 10-mm thick cyst wall. The inner wall of the cyst was yellowish and appeared granular. No atypia or malignancy was noted in the omental biopsy nor in the cytology of the peritoneal fluid taken.

Postoperative recovery was uneventful. The patient remained asymptomatic with no recurrence 30 months after the operation.

Discussion

Xanthogranulomatous cystitis may present with urinary symptoms, abdominal pain or umbilical discharge.² This condition is characterised microscopically by multinucleated giant cells, lipid-laden macrophages (xanthoma cells) and cholesterol crystals.^{3,4} Macroscopically, it manifests as soft yellow-brown plaques.⁴ 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 characterised by basophilic lamellar inclusion bodies (Michaelis Gutmann bodies) and large aggregates of monocytes (Von Hanseman bodies).⁵

Twenty-three cases have been reported in literature, with the following clinical characteristics: median age of 45 years (range, 16 to 76 years old); no obvious sexual predilection (12 of 23 cases were female, 1 case of unknown age and sex); 11 patients were Japanese; the majority of lesions were located in the bladder dome (18 of 21 cases or 85.7%,

of these 3 cases were of unknown location) and mostly associated with a urachal remnant (17 of 24 cases or 70.8%).

The aetiology of xanthogranulomatous cystitis is unclear. Proposed explanations suggest a chronic inflammatory process caused by mechanisms such as (i) immunological defect of the macrophage,³ (ii) chronic infection of the urachal diverticulum or cyst,⁶ (iii) gram negative or anaerobic bacteria such as in urinary tract infections⁵ or infection after tubal ligation,¹ (iv) foreign material such as retained suture material,⁶ (v) local response to a bladder tumour,³ and (vi) abnormal lipid metabolism and lipid accumulation in a macrophage.^{4,7} In our patient, previous tubal ligation with local inflammation could be a predisposing cause.

Diagnosis is difficult preoperatively and the condition may be mistaken for a malignancy. Xanthogranulomatous cystitis may also be associated with carcinoma:³ this dual pathology should be sought during surgical exploration. Although uncommon, urachal carcinomas must be considered due to their association with the urachal remnant. Urachal carcinomas have a male predilection and prognosis is poor, typically presenting with local invasion or metastases.

Surgical resection is curative with no postoperative recurrence reported.⁵ Simple excision may suffice for a localised xanthogranuloma but if the disease is combined with an urachal remnant or adenoma, partial cystectomy is preferred. More extensive surgery, including excision of the urachal tract is needed if urachal carcinomas are suspected. Additional antibiotic therapy may be considered in cases with positive microbial cultures.¹ However, since most patients have an associated urachal remnant as a source of chronic bacterial infection, conservative treatment is considered unwise.²

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