Xanthogranulomatous Salpingitis: A Case Report with Radiological and Histopathological Features

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Abstract

Xanthogranulomatous inflammation is a rare form of chronic destructive inflammatory process affecting different organs in the human body. Xanthogranulomatous salpingitis is a relatively rare entity, and to date a limited number of cases have been reported in the literature. Its clinical and imaging features are confusing and can mimic various benign and malignant adnexal masses. We present such a case in a 54-year-old female with uterine leiomyoma, hypertension, type II diabetes, history of lower segment caesarean section and myomectomy who presented with one week history of lower abdominal pain. Initial CT scan showed a large, complex, multilocular cystic right adnexal mass with surrounding inflammatory changes and multiple cystic para-aortic lymph nodes. This was thought to represent a tubo-ovarian abscess or mucinous neoplasm of the right ovary. It was further evaluated by a pelvis MRI which showed similar findings. The patient underwent midline laparotomy followed by total abdominal hysterectomy, bilateral salpingo-oophorectomy, total omentectomy, appendectomy and para-aortic lymph node dissection for a presumed mucinous ovarian neoplasm. Histopathological examination identified the condition as chronic xanthogranulomatous salpingitis. Although rare, xanthogranulomatous salpingitis should be kept in mind as a differential diagnosis of adnexal tumor and a pre-operative diagnosis should be sought to avoid radical surgical treatment, especially in young patients.

Keywords: Xanthogranulomatous inflammation, Xanthogranulomatous Salpingitis, mucinous cystic adenocarcinoma, radiology

Introduction

Xanthogranulomatous inflammation (XGI) represents a chronic inflammatory process that detrimentally impacts the normal tissue of the affected organs. It is defined by the abundance of macrophages laden with lipids, accompanied by a mix of lymphocytes, plasma cells and neutrophils along with multinucleated giant cells.[1-3] XGI can affect various organs, with the kidney being the most frequently affected.[4] The occurrence of this disease in the female genital tract is extremely rare, with only a small number of reported cases.

Due to its ability to cause local tissue damage and form masses through adhesions, this inflammation can occasionally resemble malignancy, exhibiting similar clinical and radiological features.[5,6] Here, we report a case of xanthogranulomatous salpingitis (XGS) with characteristic imaging features that can help in differentiating it from malignancy to avoid radical surgical treatment, especially in young patients.

Case Report

A 54-year-old female was referred to our institution with one week history of worsening lower abdominal pain associated with constipation, nausea, and dysuria. Her past medical history was significant for hypertension and diabetes which were controlled on medications. She had a history of a large single fundal intramural uterine fibroid, which was treated with a myomectomy five years prior to her current presentation. She had also a cesarean section at a younger age. She was afebrile and hemodynamically normal. Physical examination showed abdominal distension with tenderness and guarding in the right lower abdomen. The laboratory values were; Haemoglobin: 11.70 g/dL, WBC: 21.1 x 10^3/uL, Neutrophil: 18.2 x 10^3/uL, CRP: 141 mg/L, CA- 125: 314 U/mL. Other blood tests and tumor markers (B-HCG, CA 19-9, CEA, AFP, LDH) were normal. A urine culture showed no growth.

Contrast-enhanced abdominopelvic computed tomography (CT) was done. It demonstrated a large 12-cm, multiloculated right adnexal cystic mass with thick enhancing walls and significant surrounding fat stranding, soft tissue thickening, and omental nodularity. The mass was inseparable from the adjacent anterior abdominal wall and uterine wall. There was significant reactive circumferential terminal ileal and caecal wall thickening with tethering of the bowel loops and mesentery. The left ovary showed rim enhancing cystic lesion measuring 2.2 cm. Additionally, multiple enlarged low-attenuation/cystic para-aortic lymph nodes were seen, and the CT findings were initially thought to represent possible tubo-ovarian abscess or localized rupture of mucinous right ovarian tumor. Tuberculosis was considered given low-attenuation retroperitoneal lymph nodes (Figure 1).

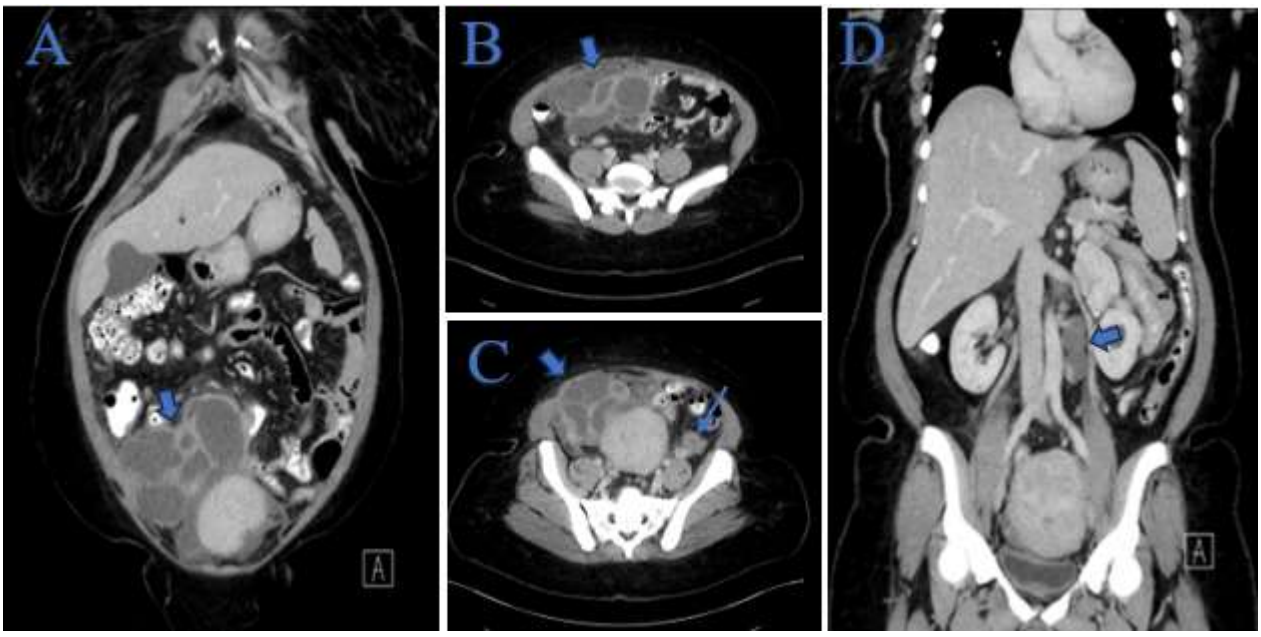

Figure 1: Computed Tomography (CT) with IV contrast in the portal venous phase images. (A) Coronal image, (B) and (C) Axial images showing multilocular right adnexa cystic mass with thick enhancing walls and significant surrounding fat stranding, soft tissue thickening, and omental nodularity (short solid arrows), (C) The left ovary showed rim enhancing cystic lesion measuring 2.2 cm (long solid arrow). (D) Coronal image showing enlarged left para-aortic lymph nodes with cystic area suggesting necrosis (short solid arrow).

A pelvis MRI was subsequently performed which confirmed the presence of a large 11-cm right pelvic complex multilocular cystic mass. Some of the locules were interconnected to each other forming a tubular structure. Different cystic locules contained different T2 signal intensities giving a stained-glass appearance. The areas with low signal intensity on T2-weighted images demonstrated diffusion restriction and post contrast images showed enhancement of the thickened walls and septae. Perilesional inflammatory changes were noted, evident as hyperintense signals of the surrounding fat on T2-weighted images. There was T2 hyperintense fluid in the pelvis anteriorly and in the left adnexa and the distal ileal loops were adherent to the mass with reactive thickening, enhancement, and congestion of the

vessels. The cystic mass was inseparable from the right ovary and fallopian tube and was favoured to be ovarian in origin. The presence of pelvic free fluid, omental nodularity and nodular peritoneal thickening was thought to represent metastatic disease with possible rupture of the ovarian complex cystic mass. Furthermore, the presence of contralateral left ovarian cystic lesion and cystic para-aortic lymph nodes were deemed metastatic in origin. Considering the presence of lymphadenopathy, the diagnosis of mucinous cystadenocarcinoma was favoured (Figure 2).

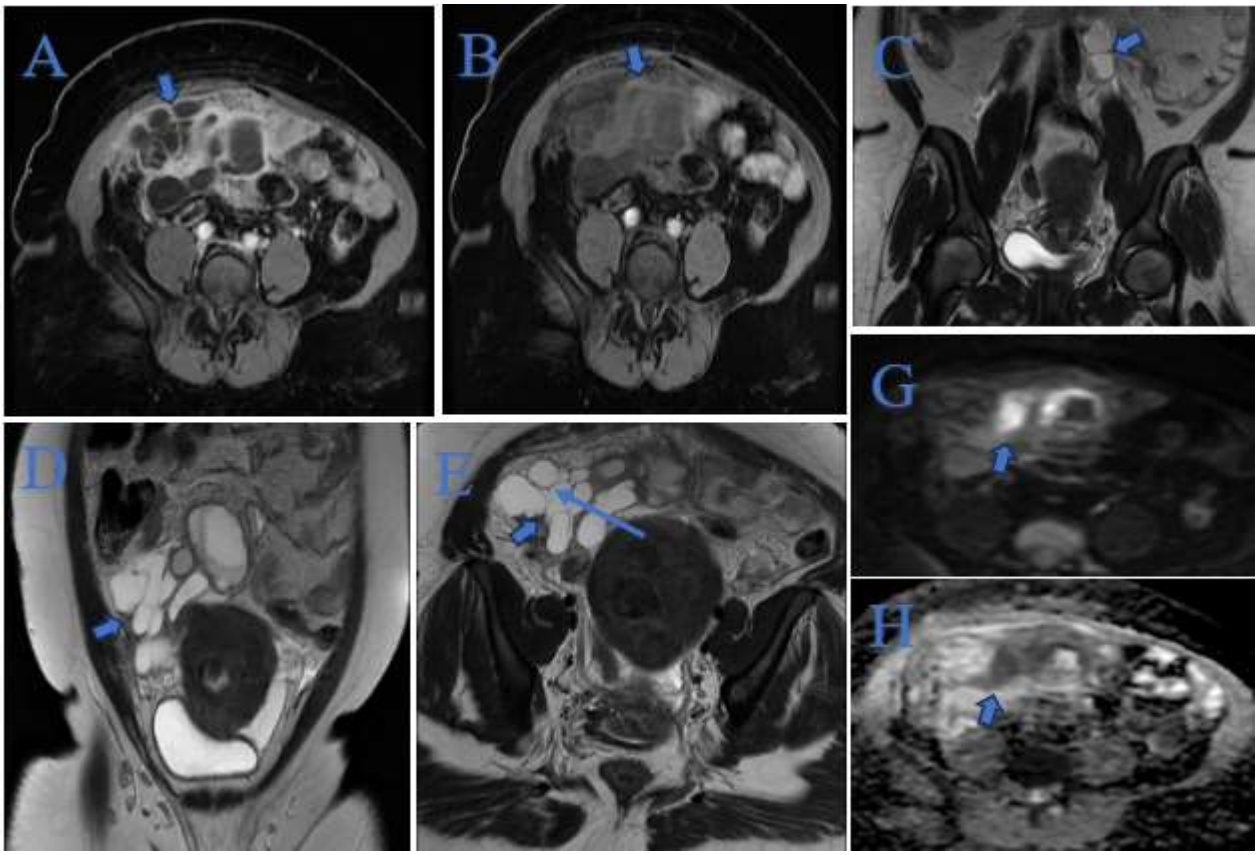

Figure 2: Magnetic Resonance Imaging (MRI) images. (A) Axial T1WI post contrast venous phase showing a large multilocular cystic mass with thick enhancement of the walls and septations with irregular margins (solid short arrow). (B) Axial T1WI without IV contrast showing hyperintense walls of the cystic mass (solid short arrow). (C) Coronal T2WI showing cystic lesions in the left para-aortic region correspond to previously seen para-aortic lymph nodes (solid short arrow). (D) Coronal and (E) Axial T2WI showing complex multilocular cystic mass and some of the locules interconnected to each other forming tubular structures with variable signal intensity of these locules (solid short arrow) and significant surrounding inflammatory changes with edema manifested by hyperintense signals of the surrounding fat (solid long arrow). (G) Axial DWI showing region of high signal intensity which demonstrates diffusion restriction in ADC image (H).

Ultrasound-guided aspiration of the cyst contents was performed and the cytology of collected fluid showed neutrophilia with few lymphocytes, and macrophages.

The patient underwent midline laparotomy and extensive debulking surgery which included total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy, appendectomy, and para-aortic lymph nodes dissection. Intra-operative findings were of a mass attached to the right ovary and ileum measuring 4 x 3 cm with gross ascites and multiple enlarged para-aortic lymph nodes. Microscopic examination of the collected peritoneal fluid was negative for malignancy.

Post-operative histological examination revealed a grossly swollen right fallopian tube with two para-tubal cysts. Microscopic examination showed dense infiltration of the right fallopian tube by lymphoplasmacytic cells admixed with sheets of histiocytes in the tubal plicae as well as in the wall. The underlying fibro-collagenous stroma showed marked congestion and a section from the collected friable mass showed fibroadipose tissue with dense infiltration of sheets of histiocytes. There was edema and congestion along with lymphoid aggregates on the surface of the right ovary and in the omentum. Small bowel seedings were negative for atypia or malignancy and showed only Histiocytic reaction. Examined lymph nodes showed reactive changes and some showed Mullerian inclusions. The Immunohistochemistry of these cells showed positive CD68 confirming the diagnosis of chronic Xanthogranulomatous Salpingitis (Figure 3).

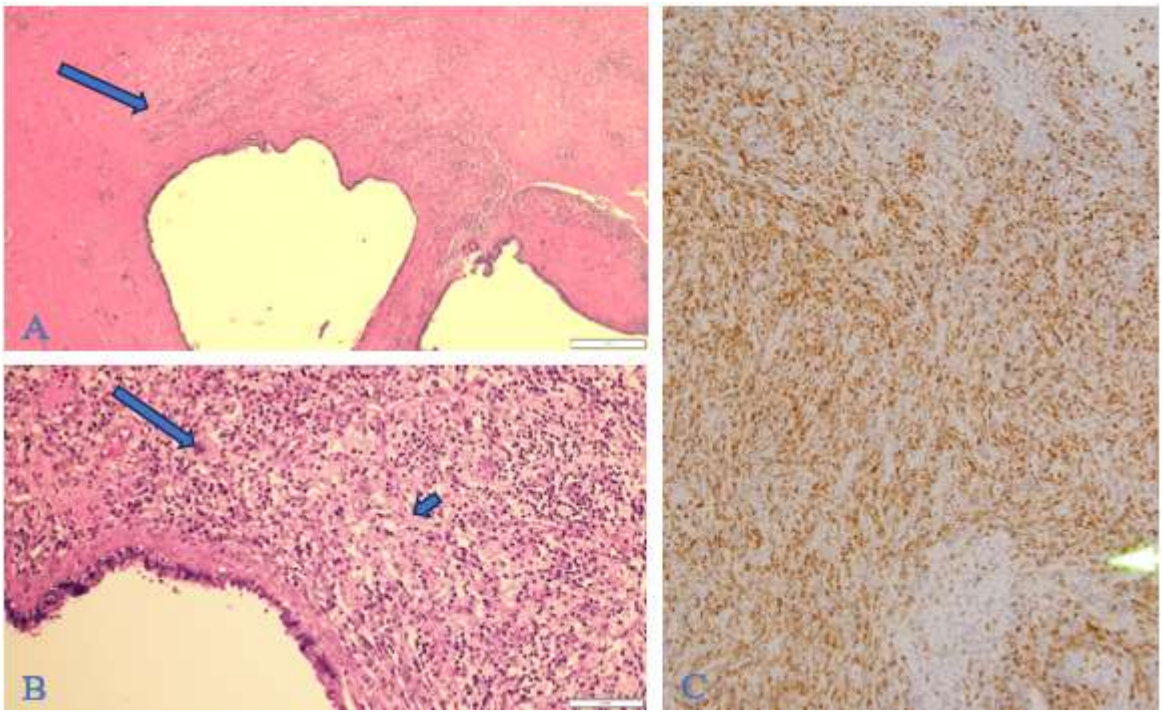

Figure 3: (A) Low power view (4x) shows fallopian tube epithelium surrounded by dense inflammation (solid long arrow). (B) High power view (40x) shows diffuse infiltration by lipid laden macrophages (solid long arrow) admixed with few multi-nucleated giant cells (solid short arrow).(C) shows intermediated power view showing positive CD68 immunostain in histiocytes.

Discussion

Xanthogranulomatous inflammation of the female genital tract is extremely rare. It was initially identified by Kunakemakron et al. in 1976.[7] Since this discovery, approximately a dozen cases of xanthogranulomatous salpingitis have been reported, mostly as case reports including a recent case series of thirteen patients.[8] Few other cases of xanthogranulomatous inflammation affecting both the fallopian tube and ovary have been reported.

In our case, the patient presented with lower abdominal pain and blood test showed high inflammatory markers. In previous literatures, xanthogranulomatous inflammation has been documented across a wide age range, typically reported between 23 to 72 years old.[9,10] Yet, Tanwar et al. reported a distinct case of xanthogranulomatous salpingo-oophoritis in a two-year-old girl who presented with an abdominal mass.[11] The clinical presentations vary, with the most frequent ones being an abdominal mass, abdominal pain, and vaginal bleeding.[12] The pathogenesis of XGS is uncertain; several theories have been suggested, including infection, endometriosis, contraceptives, lipid metabolism issues, drugs and presence of leiomyoma[10] as found in our case.

Although MRI findings of xanthogranulomatous pyelonephritis and cholecystitis have been described in the literature[13,14] and about two case reports of xanthogranulomatous oophoritis included detailed radiological findings,[15,16] there is no published data about imaging description of xanthogranulomatous salpingitis. Most of the previously reported cases showed an adnexal mass with cystic component[2,3,6,11,17-19] as in our case. In the two reported cases of xanthogranulomatous oophoritis,[15,16] MRI findings displayed a multiloculated cystic mass with thickened, enhancing walls and different signal intensity in each locule, which is similar to the findings in our patient. However, these cases also showed multiple non-enhancing nodules within a thickened wall in T2-weighted images, a feature noted in xanthogranulomatous cholecystitis[13,14] but absent in our case. Several distinctive radiological features found in our case can aid in the pre-operative diagnosis of xanthogranulomatous salpingitis and spare unnecessary extensive surgery. Firstly, the tubular appearance of the identified cystic mass indicates it likely originates from the fallopian tube, though it could also signify a twisted fallopian tube as a complication of an ovarian mass. Secondly, the confinement of inflammatory changes (omental fat stranding, peritoneal nodularity and free fluid) to the area adjacent to the mass with sparing of the other areas is possibly helpful.[6] On the other hand, metastatic deposits typically spread across the abdomen and pelvis reaching up to the level of the diaphragm. Thirdly, in MRI, the cyst contents showed a high-signal intensity on Diffusion-weighted imaging (DWI) with a low apparent diffusion coefficient (ADC) map which is reminiscence of DWI findings of xanthogranulomatous Oophritis.[15] In our case, diffusion restriction was observed inside the cystic lesion, not within its walls as typically observed in malignancies. Finally, presence of enlarged lymph nodes was not addressed in the previous reported cases. Interestingly, those lymph nodes were almost completely cystic with small solid component/mural thickening. In our case, the imaging characteristics of the mass were thought to represent mucinous cystadenocarcinoma and the presence of enlarged lymph nodes raised the suspicion of malignancy. In summary, a clue to possible correct diagnosis of xanthogranulomatous salpingitis is presence of multiloculated cystic mass showing tubular appearance with confined inflammatory process and restricted diffusion within the cystic component rather than at the septa or walls.

Xanthogranulomatous Salpingitis is a rare condition and has solely been identified by histopathological examination until present. Given its rarity, the imaging appearances might be confusing and misleading. Medical treatment using antibiotics has not demonstrated any appreciable benefit in reducing the inflammation and complete surgical resection of the affected fallopian tube is considered to be a definitive and effective treatment approach.[3] Therefore, the knowledge of the supporting MRI imaging features of Xanthogranulomatous Salpingitis may prevent extensive radical surgical treatment.

Conclusion

In conclusion, diagnosis of Xanthogranulomatous Salpingitis is challenging due to its rarity. This case report aimed to emphasize the important radiological findings that could assist in diagnosing the condition before surgery and elaborating on the typical histopathological findings.

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