

Xanthogranulomatous Oophoritis Masquerading as Malignant Ovarian Tumor: A Case Report

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ABSTRACT

Xanthogranulomatous inflammation of the genital tract is uncommon but usually involves the endometrium. A 19-year-old girl presented with an abdominopelvic lump with a history of fever, irregular menses, and loss of appetite. On examination, a cystic, irregular, mobile mass of around 22-24 weeks' uterine size was felt. On investigation, CA-125 was 16 U/mL, CA19.9 was 2.43U/mL, AFP was 0, and Inhibin B was 319.92 pg/mL. The CT scan mimicked ovarian malignancy. She underwent an exploratory laparotomy for the same reason, but upon rupture, cheesy material appeared from the mass. A frozen section was sent with pus culture showed no growth, and the pus culture showed no growth. However, the final histopathology report suggested foamy macrophages, neutrophils, and CD68-positive histiocytes, indicative of xanthogranulomatous oophoritis.

Such patients present with a long-standing history of pelvic inflammatory disease and symptoms such as anorexia, fever, and lower abdominal pain. The radiological findings mimic ovarian malignancy. A frozen section can be helpful to rule out malignancy. Xanthogranulomatous oophoritis is a rare entity that poses a diagnostic challenge.

Keywords: Dilemma; Malignant ovarian tumor; Ovarian tumors; Oophoritis; Xanthogranulomatous

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Introduction

Xanthogranulomatous inflammation is an exceptional, benign, chronic inflammatory pathology that destroys the normal tissue of organs such as the kidney, gallbladder, alimentary canal, bone, urinary bladder, testis, and epididymis through lipid-containing inflammatory cells. Involvement in the female genital tract is atypical. However, when present, it is mostly restricted to the endometrium. Cases affecting the vagina, cervix, fallopian tube, and ovary have also been re-

ported (1). For the first time in 1976, Kunakemakorn diagnosed xanthogranulomatous inflammation of the ovary and labeled it as a pseudotumor of the ovary, comparable to malignancies. Sparse data in the literature have been reported; thus, this case report has been brought to light (2).

Case Report


A 19-year-old, unmarried girl presented to the outpatient department of our hospital with a mass per abdomen for 6 months, abdominal pain for 2 months, and low-grade fever for 15 days, for which she has been taking over-the-counter medications for fever and pain. She had a history of loss of appetite and weight loss. Her menstrual cycle had been irregular for the last 3 months, occurring every 35-40 days and lasting 4-5 days, with 3-4 pads soaked per day. It was also associated with severe dysmenorrhea and occasional passage of clots.. She had no history of contact with tuberculosis. On general examination, the girl was underweight with a BMI of 18 kg/m², afebrile, and her vitals were stable with pallor 1+. On abdominal examination, an abdomino-pelvic lump of approximately 22-24 weeks' uterine size with irregular borders, smooth surface, non-tender, mobile from side to side, and cystic in consistency was felt, with the lower margin not being reachable. Further investigation revealed that USG suggested a normal-sized uterus with an ET of 5mm and a complex, multiloculated, and multiseptated (thick and vascular) cystic lesion of 10×13×12 cm, possibly neoplastic, arising from the

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right ovary, with right hydrosalpinx and suspicious omental thickening (Figure 1 and Figure 2). The CECT of the whole abdomen revealed a large, multiseptated, multiloculated solid-cystic lesion measuring 13.2×11.1×14.3 cm, abutting the bladder and abdominal wall, arising from the right ovary. A similar lesion of 4.8×2.5 cm was seen in the left adnexa, with inflammatory fat strands in the pelvis. The lesion caused right hydroureteronephrosis and reactive retroperitoneal lymph nodes. The Mantoux test was negative. Tumor markers were reported as CA-125 was 16 U/ml, CA19.9 was 2.43 U/mL, AFP was 0, Inhibin B was 319.92 pg/ml, LDH was 250 IU/L, and TSH was 0.81 uIU/mL, with the rest of the blood parameters within normal limits..



Figure 1: USG appearance of ovarian cyst

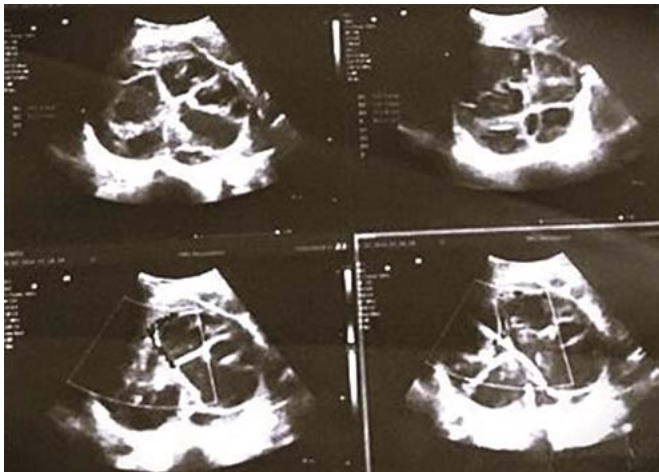


Figure 2: USG appearance of ovarian cyst

She was planned for exploratory laparotomy and proceeded. Intraoperatively, a mass of 14×12 cm was seen arising from the right adnexa, adhering to the anterior abdominal wall, lateral pelvic wall, and fundus of the bladder with necrosed tissue present throughout the pelvis. A frozen section was sent and reported as ovarian malignancy. Adhesiolysis was performed and the mass was ruptured iatrogenically with pus-like cheesy material draining from the mass (sent for culture and sensitivity). All the necrotic tissue present encasing

the uterus and bladder was excised and sent for histopathology (Figure 3 and Figure 4). Infracolic omentectomy and right pelvic lymph node dissection were done for complete staging and sent for histopathology. The uterus with left adnexa and the rest of the abdominal solid organs were normal. She was managed postoperatively with high-dose antibiotics and adequate analgesia. She recovered well and was discharged. On follow-up, the pus culture report from the mass was sterile, and histopathology was reported as solid areas with foamy histiocytes, plasma cells, and lymphocytes, along with inflammatory cells in the mass, omentum, and necrotic areas. The lymph nodes showed reactive hyperplasia and immunohistochemistry of foamy histiocytes with CD68 positivity, indicative of xanthogranulomatous oophoritis.

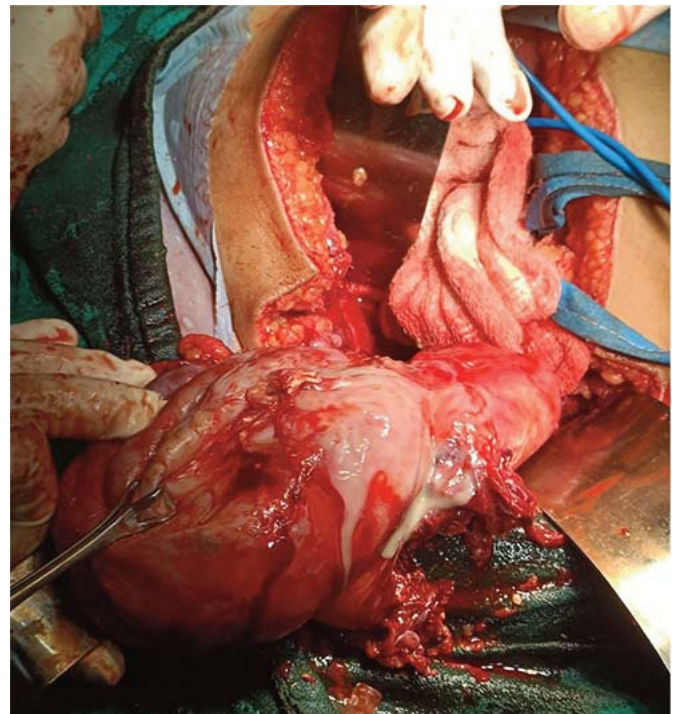


Figure 3: Intra-op appearance of xanthogranulomatous infection of this patient

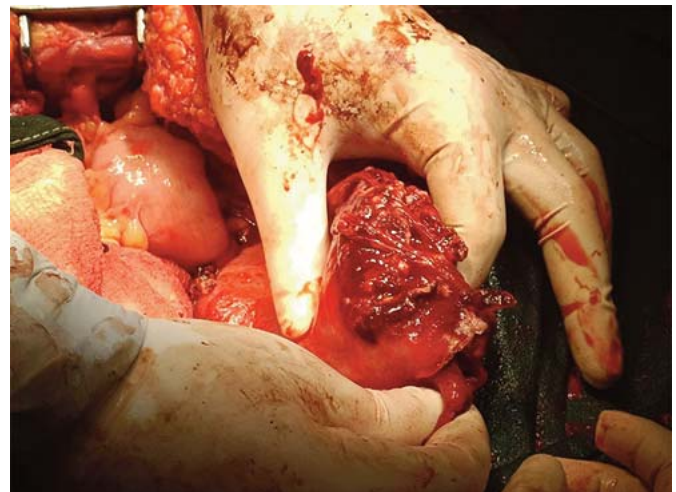


Figure 4: Intra-op appearance of necrosed tissue over the bladder of this patient

Discussion

Xanthogranulomatous oophoritis usually affects females of age group 2-84 years with no exact etiology, yet various conjectures have suggested its correlation with leiomyoma, infections such as tuberculosis, endometriosis, and Crohn's disease. Additionally, it has been associated with the use of Intrauterine Contraceptive Devices (IUCD), inappropriate drug intake, defects in lipid metabolism, foreign body reaction to suture material, necrotizing reaction introduced during previous operative procedures, previous diathermy, and bacterial tubo-ovarian abscess (3-5). Infection is the most recognized theory, upheld by proof of the presence of organisms such as *Escherichia coli*, *Bacteroides fragilis*, *Proteus vulgaris*, *Staphylococcus* spp., and *Salmonella typhi* in the tissue, as shown by culture and sensitivity reports (5). Theories suggest that the IUCD in the endometrial cavity leads to bacterial shedding in the fallopian tube. During ovulation, the ovarian surface gets exposed to these bacteria, which causes the corpus luteum to become infected, ultimately leading to the formation of an ovarian abscess. (6).

It poses a diagnostic and therapeutic challenge for the clinician due to the involvement of surrounding structures. The usual symptoms and signs of this disease consist of lower abdominal or suprapubic pain, fever, anemia, menorrhagia, and adnexal tenderness. During a pelvic examination, an adnexal mass may be palpated with or without tenderness (7,8). Laboratory tests show elevated ESR and raised white blood cell count (9). In this case, the patient had a history of fever and abdominal pain, which was managed with over-the-counter medications.

A mass lesion in the pelvic cavity encroaches on adjacent tissue, impersonating a neoplastic lesion as observed radiologically. Imaging studies, such as USG, may reveal adnexal masses suspicious for ovarian neoplasia, which may be solid to cystic, nodular, with inflammation, and may even extend beyond the ovaries, encompassing adjacent organs. A CT scan may reveal a well-defined solid lesion with altered signal intensity, mistakenly identified as ovarian malignancy (2,10).

In our case, the differential diagnosis was ovarian malignancy or tuberculosis on the basis of clinical findings and CT scan reports. On MRI, it is observed as a high-signal-intensity mass with multiple low-signal-intensity internal septations on axial T2W turbo spin echo. In addition, irregularly thickened walls with hyperintense nodules clustered in the ovarian mass have also been reported. The differential diagnosis of such cases usually includes endometriosis, tuberculosis, or fungal infection and can be ruled out by specific culture reports and special stains (11,12). Six cases of different age groups (mean age of 22 years) have been clinicoradiologically misinterpreted as malignant ovarian neoplasm in the past during a study period of five years. All these six patients presented with abdominal pain and adnexal mass; half of them had fever and weight

loss. On the USG and CECT abdomen, all had an adnexal mass on one or both sides, with or without extension into surrounding structures. All patients were operated on for suspected neoplastic disease, and histopathology was confirmatory for the diagnosis of xanthogranulomatous infection (5).

The preferred treatment for xanthogranulomatous oophoritis is oophorectomy; however, due to the diagnostic dilemma, extensive surgery is frequently performed (2,4,6). The intraoperative practice of frozen section aids in reducing the surgical burden on the patient. In this case, it was reported as malignant (8). Pus culture in this case was found to be sterile. On gross examination of such specimens, the involved ovary has a well-circumscribed solid to cystic, yellowish, lobulated mass with prominent microscopic infiltration of lipid-laden macrophages, known as xanthoma cells, and inflammatory cells consisting of lymphocytes and plasma cells along with a few neutrophils (13). Kishore et al. suggested malakoplakia as a key differential diagnosis owing to the presence of foam cells, and it is distinguished from xanthogranulomatous oophoritis by basophilic Michaelis-Gutmann bodies, which are absent in the latter (7). Immunohistochemical stains CD68, CD3, and CD20 are also helpful in establishing a diagnosis (9). However, they are seldom used due to their unique histopathological characteristics, except in this case.

Declarations

Ethics: Informed consent was obtained from the patient before publishing this article.

Availability of data and materials: The data supporting this study is available through the corresponding author upon reasonable request.

Competing interests: The authors declare that they have no competing interests.

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Authors' contributions: SG, and SKA: Raised and designed the presented report. SKA and SR: Developed the first draft of the manuscript. All authors contributed to the writing of the paper, and have read and approved the final manuscript. SR: Assisted with data collection and analysis. SG: Critically revised the manuscript. All authors read and approved the final manuscript.

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