CASE REPORT

Xanthogranulomatous Oophritis Masquerading as Ovarian Neoplasm: A case report

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ABSTRACT

Xanthogranulomatous inflammation is a special form of chronic inflammation that destroys normal tissue of affected organs which get replaced by chronic inflammatory cell infiltrate admixed with foci or sheets of foam cells, fibroplasias and vascular proliferation. Only a few cases of xanthogranulomatous oophoritis have been reported. We report a case of 35 year old female who presented with lower abdominal pain and polymenorrhea. A total hysterectomy with bilateral salpingoopherectomy was performed on the suspicion of an ovarian neoplasm.

Keywords: Ovary, neoplasm, xanthogranulomatous, oophritis

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Xanthogranulomatous inflammation is a special form of chronic inflammation that is destructive to normal tissue of affected organs.¹ It is characterized by the presence of a large number of lipid-containing macrophages with an admixture of lymphocytes, plasma cells and neutrophils.² Most commonly affected organ is kidney followed by gallbladder.³ Female genital tract is an unusual location for xanthogranulomatous inflammation and if present usually involves the endometrium. Only a few cases of xanthogranulomatous oophoritis have been reported^{4,5}

CASE REPORT

A 35-year-old woman originally presented with lower abdominal pain and polymenorrhea along with intermittent fever since three months. She had no co existing medical or surgical co-morbidity. On examination, a large mass filling the right pelvic cavity was noted. CA-125 levels were elevated (171.4) in the patient.

She was evaluated for the pelvic mass and a CT scan performed revealed a large 5.8 x 4.3 cm complex solid cystic mass with multiple enhancing septations in the right adnexa. No calcifications were seen. Left adnexa showed a 3x2 cm follicular cyst. Few retroperitoneal nodes were also

noted.

In view of the above findings an ovarian neoplasm was suspected and hysterectomy along with bilateral salpingoopherectomy was performed. Intraoperatively a right adnexal mass adherent to the uterus and adjacent peritoneum was found. Minimal peritoneal and pelvic nodes were seen.

We received a specimen of uterus along with right tuboovarian mass and left fallopian tube with ovary along with omental fat and pelvic nodes. The mass measured 9x8x4 cm having an irregular external surface and was adherent to the uterus. Cut section was greyish white, firm along with focal cystic and necrotic areas. Left ovary was edematous measuring 5x4x3 cm. Cut section showed a 2x2 cm follicular cyst. Uterus measured 8.5x4x3 cm and cut section was unremarkable. (Fig 1)

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Fig 1: Right ovary shows the mass- grayish white, firm along with focal cystic and necrotic areas. Left ovary shows follicular cyst

Histologically, right ovarian stroma was almost completely replaced with dense inflammatory cell infiltrate consisting of large number of lipid laden macrophages, accompanied by lymphocytes and plasma cells along with areas of necrosis. Fibrosis along with vascular proliferation was also present. There was no evidence of malignancy in the sections studied from the tubo-ovarian mass. Sections from the left ovary, left fallopian tube and uterus were unremarkable. (Fig 2 & 3)

Fig 2 & 3: Ovarian stroma almost completely replaced with lipid laden macrophages, accompanied by lymphocytes and plasma cells along with fibrosis and vascular proliferation. (H and E X 400)

DISCUSSION

Xanthogranulomatous inflammation is a rare clinical condition. However, when encountered it occurs commonly in the lung, orbit, renal and biliary system³. It is a special form of chronic inflammation that is destructive to normal tissue of affected organs¹. Xanthogranulomatous inflammation occurring in female genital tract affects endometrium, fallopian tubes or ovaries focally or entirely, which clinically forms mass like lesion in the pelvic cavity and invades the surrounding tissues⁶. Kunakemakorn was the first to describe xanthogranulomatous inflammation of serosa of uterus, left fallopian tube and ovary in his report of inflammatory pseudotumor in the pelvis in 1976⁷. Only 16 cases of xanthogranulomatous inflammation involving the ovary have been reported until date with very few from Indian literature⁸. The clinical picture, radiological findings and the gross findings in a xanthogranulomatous ovary are amenable to confusion with an ovarian malignancy.

The etiopathogenesis of this condition has been debated over years and is still unclear. Proposed risk factors include chronic bacterial infections, ineffective antibiotic therapy, use of IUCD, abnormalities in lipid metabolism, endometriosis, foreign material such as retained suture material⁹. Our patient had been on an antibiotic therapy for a while and was also using IUCD. Bacteria like *Bacteroides fragilis*, *Proteus*, *Escherichia coli*, *Staphylococcus aureus*, *Salmonella typhi* can be considered in the pathogenesis of xanthogranulomatous oophoritis¹⁰. Punia et al reported xanthogranulomatous oophoritis as a late sequelae of inadequately treated pelvic inflammatory disease¹¹. Singh et al reported a case 3 yrs following uterine artery embolisation¹². U R Singh et al reported xanthogranulomatous oophoritis as a complication of typhoid¹³.

The age group of patients in previously reported cases varied mostly from 17 to 73 years^{13,14,15} with the youngest case reported in a 2 yrs old female in 2015¹⁵. The clinical presentation include fever, abdominal pain, menorrhagia, abdominal mass, anemia. The involved ovary in each of the previous reported cases was replaced by solid, yellow, lobulated masses that was well circumscribed and showed xanthogranulomatous inflammation on microscopy^{16,17}.

USG may reveal a solid-cystic mass with septations and

clear margins. A CT scan may reveal a well-defined solid mass with altered signal intensity⁴. Presence of nonenhancing intramural nodules in the thickened wall of an ovarian cystic mass with high signal intensity on T2-weighted images and low signal intensity on T1-weighted images may be a unique MR indicator of xanthogranulomatous oophoritis^{18,19}. CT and MR findings are however usually nonspecific for this condition as they are frequently misdiagnosed as malignant ovarian tumors due to appearances unusual for tuboovarian abscesses²⁰.

The condition needs to be differentiated from various neoplastic and non neoplastic conditions. The differential diagnosis includes non neoplastic conditions like tuberculosis, fungal infections, malakoplakia and neoplastic conditions like lymphoma, leukaemia, sclerosing stromal tumor and malignant small round cell tumor. Definitive diagnosis, however, can only be made on microscopy. Morphologically, the normal ovarian structure is destroyed and replaced by chronic inflammatory cell infiltrate admixed with foci or sheets of foam cells, fibroplasia, chronic inflammatory cell infiltrate and vascular proliferation similar to the picture seen in our case. The pathologist however has to be very cautious as a mild increase in any of these components can lead to misdiagnosis. Immunohistochemistry for CD68, Vimentin, S100 are helpful^{4,5} in confirming the diagnosis.

The treatment of choice for xanthogranulomatous oophoritis is oophorectomy. Antibiotics have been attempted but they have been unsuccessful in reducing the size of the mass⁷. Since this is associated with many other conditions like pelvic inflammatory diseases, endometriosis and intrauterine death, the patient needs to be kept on a close follow up.

CONCLUSION

Xanthogranulomatous oophoritis is a rare benign condition with very few cases reported in the literature. It can be easily misinterpreted as malignancy by the clinical picture and various imaging modalities making histopathology the most definitive method of diagnosing the condition. Also, patients on long term antibiotics, endometriosis and pelvic inflammatory diseases need to be kept on a close follow up as they are more prone to get xanthogranulomatous oophoritis.

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