Case Report

Xanthogranulomatous Oophoritis Mimicking an Ovarian Neoplasm: A Rare Case Report

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Abstract

Xanthogranulomatous inflammation of female genital tract is uncommon and is usually seen in endometrium. Only a few cases involving the ovary have been reported. Its clinical manifestations, imaging modalities, and gross features can mimic ovarian malignancy. Thus, a preoperative diagnosis of this entity is important to avoid radical surgical treatment. We report a case of xanthogranulomatous oophoritis in a 24 years female, which was clinically and radiologically misdiagnosed as ovarian neoplasm. The approach to such a case and the differential diagnosis has also been discussed.

Keywords: *Oophoritis, ovarian, xanthogranulomatous*

Introduction

Xanthogranulomatous inflammation is an unusual type of chronic inflammation that leads to tissue destruction by inflammatory cells comprised of lipid-containing macrophages, lymphocytes, plasma cells, multinucleated giant cells, and neutrophils. Xanthogranulomatous inflammation of female genital tract is uncommon and is usually seen in endometrium. Only a few cases involving the ovary have been reported.[1] Its presentation as a mass lesion in pelvic cavity and invasion of surrounding tissue can mimic a neoplastic lesion clinically, radiologically, and on gross examination.

We report a case of xanthogranulomatous oophoritis in a 24-year-old female, which was clinically and radiologically misdiagnosed as ovarian neoplasm. Histopathological findings revealed characteristics features of xanthogranulomatous oophoritis.

Case Report

A 24-year-old $P_3L_3A_3$ female presented with the chief complaints of pain abdomen on and off, and heavy irregular cycles for 6 months. On per vaginal examination, a firm left adnexal mass could be palpated. Her blood investigations revealed microcytic hypochromic anemia, slight raise in total leukocyte count (12,000/L), raised erythrocyte sedimentation rate (ESR) (45 mm at 1st h), and slightly raised lactate dehydrogenase (490 IU/L). Mantoux test was negative. Contrast-enhanced computed tomography abdomen was performed, which showed a left adnexal mass, measuring 8.5 cm × 6.8 cm × 6.3 cm, causing displacement of surrounding bowel loops and lateral displacement of uterus. The mass was multiloculated, cystic with enhancing irregular walls

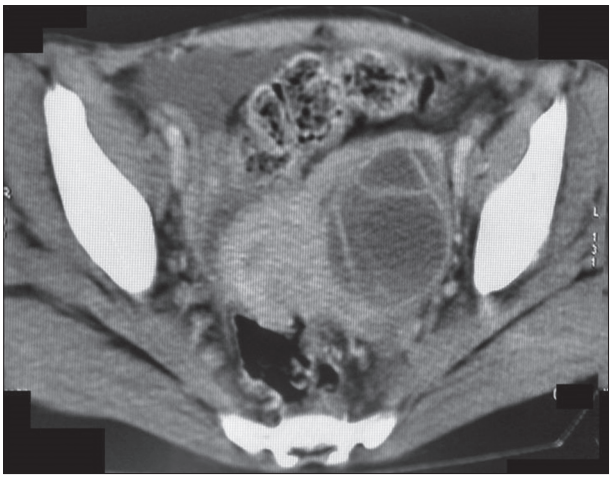

Figure 1: Contrast-enhanced computed tomography abdomen showing a left adnexal mass, which is multiloculated, cystic with enhancing irregular walls and multiple septations

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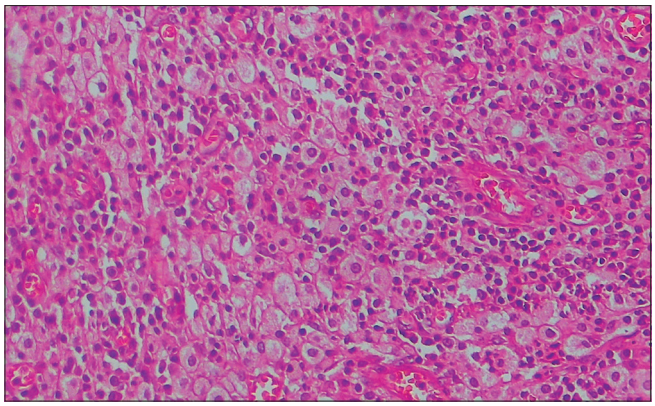

Figure 2: Section from tubo-ovarian mass showing dense granulation tissue formation with abundant foamy macrophages, along with lymphoplasmacytic cells and congested blood vessels (H and E, ×10)

and multiple septations [Figure 1]. The possibility of a benign neoplasm (cystadenoma) was suggested.

An exploratory laparotomy followed by left salpingo-oophorectomy with pus drainage followed by right tubal ligation was performed. Grossly, the tubo-ovarian mass had a pearly white capsule. On cut, it was gray-white with multiple solid cystic areas, with cysts varying from 0.2 to 3 cm in diameter. On microscopy, a solid cystic lesion was seen, with normal ovarian tissue at the periphery. There was dense granulation tissue formation with abundant foamy macrophages, surrounded by fibrocollagenous tissue showing infiltration by lymphoplasmacytic cells and macrophages, along with congested blood vessels [Figure 2]. Thus, a final diagnosis of xanthogranulomatous oophoritis was given.

Discussion

The female genital tract is very rarely affected by xanthogranulomatous inflammation, and usually, it is confined to the endometrium. Only a few cases of xanthogranulomatous oophoritis have been reported from India.[1]

The average age of patients with affected ovaries is 31 years,[2,3] and the youngest case reported was of 2 years old. The clinical presentations include pain in abdomen, fever, abdominal mass, menorrhagia, anemia, and anorexia. Gynecological examination reveals adnexal mass with tenderness. Laboratory tests show elevated ESR and raised white blood cell count. Radiological findings of xanthogranulomatous oophoritis may simulate a malignant ovarian neoplasm, due to the involvement of adjacent organs and pelvic peritoneum resulting in adhesions. Grossly, the involved ovary is enlarged and replaced by a solid, yellow lobulated well-circumscribed mass, sometimes involving adjacent organs, thereby mimicking malignancy.[1] Microscopically, there is infiltration of sheets of foamy cells admixed with mixture of inflammatory cells such as lymphocytes, plasma cells, neutrophils with or without multinucleated giant cells.[4] Foamy histiocytes (xanthoma cells) are histiocytes with abundant lipid-laden cytoplasm having vacuolated appearance, responsible for the yellow color on gross examination. The emergence of foam cells may be attributed to the following factors:

1. Inefficient or inappropriate antibiotics applied in the early phase of infection that resulted in ineffective control of bacterial multiplication
2. Presence of a lipid metabolic disorder that induces hyperlipidemia and the foam cells are formed when the lipid deposited is phagocytosed by phagocytes
3. The application of intrauterine contraceptive devices or drugs.[5]

Differential diagnosis of xanthogranulomatous oophoritis includes tuberculosis and fungal infections which can be ruled out by culture and special stains for the causative organisms. Malakoplakia is one of the differential diagnoses of xanthogranulomatous inflammation, which shows cytoplasmic concentric calcific bodies (Michaelis–Gutmann bodies).

According to Karigoudar *et al*.[6] and Elahi *et al*.,[7] frozen section is helpful in the diagnosis of xanthogranulomatous oophoritis and intraoperative management, whereby unnecessary and extensive surgery can be avoided in cases of dilemma. Similarly, Chouairy *et al*.[8] are of the view that that xanthogranulomatous oophoritis may be preoperatively misdiagnosed as an adnexal neoplasm, thus necessitating frozen section to rule out malignancy, as was done in their case. Intraoperative histopathology of xanthogranulomatous oophoritis reveals chronic inflammatory tissue.

However, Pandey *et al*.[9] have suggested that xanthogranulomatous inflammation does not always masquerade malignancy, so the role of frozen section may be limited to selected cases only.

The treatment of choice for xanthogranulomatous oophoritis is oophorectomy.

In conclusion, xanthogranulomatous oophoritis is a rare lesion whose clinical manifestations, imaging modalities and gross features can mimic ovarian malignancy. Thus, a preoperative diagnosis of this entity should be considered to avoid radical surgical treatment, especially in young patients.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the

patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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