Case Report

Xanthogranulomatous oophoritis-masquerading as ovarian neoplasm: report of two cases

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Abstract

Xanthogranulomatous oophritis is an unusual chronic inflammatory condition which clinically presents as a mass like lesion in the pelvic cavity. It invades the surrounding tissue and thus mimics like a tumor. Patients with xanthogranulomatous oophoritis presents with lower abdominal or suprapubic pain, fever, menorrhagia, vaginal bleeding, adnexal tenderness and rarely as a pelvic mass. Only a few cases of this entity have been reported in India.

We report two cases of xanthogranulomatous inflammation of the female genital tract presenting with unilateral ovarian masses having clinical and radiological suspicion of an ovarian tumor. Both patients underwent panhystrectomy. Gross examination revealed enlarged unilateral ovaries with multiloculated cystic cavities on cut surface. Histopathological examination revealed massive infiltration of the ovarian tissue by lipid laden histiocytes and mixed inflammatory infiltrate suggesting a diagnosis of xanthogranulomatous oophoritis. These cases mimicked as malignancy and are of interest in view of their rarity.

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Introduction

Xanthogranulomatous inflammation is a specific type of chronic inflammatory lesion characterized by presence of large number of lipid laden histiocytes admixed with other inflammatory cells leading to destruction of the affected tissue.[1]. This lesion is documented in gall bladder, stomach, anorectal area, bone, urinary bladder, testis, epididymis, ovary, vagina and endometrium.[2] Although xanthogranulomatous inflammation is reported in the female genital tract but only few cases of ovarian involvement have been seen. Only 13 cases of xanthogranulomatous inflammation in the ovary and fallopian tube and 15 cases in the endometrium have been reported in the literatu**re.**[2,3,4,5] This inflammatory lesion shows foamy histiocytes (lipid laden), lymphocytes, plasma cells and neutrophils infiltrating and destroying the affected tissue. Xanthogranulomatous oophoritis occurs mostly in third decade with age group of 30-35 years. [3, 4] Clinical presentation of lower abdominal or suprapubic pain, pyrexia, abnormal vaginal bleeding or spotting, adnexal tenderness and pelvic mass as the common signs and symptoms observed in these patients prompting them to seek medical attention [1, 4]

We report two cases of xanthogranulomatous oophoritis in 30 and 31 year old females because of their close resemblance to malignancy and rarity

Case Report

Case 1: A 30 year old married woman presented with two months history of lower abdominal pain and fever. She had no chronic illness and on per vaginal examination a mass was felt in the right fornix leading to pushing of uterus to the left side. Her laboratory investigations revealed normal haemogram with slightly elevated CA – 125 level (436.4 U/ml). Her ultrasonographic examination showed right adnexal complex cyst with mild hepatomegaly. CECT lower abdomen revealed high density multiloculated septated cystic lesion with solid component in the right side of pelvic cavity with suspicion of malignancy. The patient underwent total abdominal hysterectomy with bilateral salpingo-opherectomy. Per operatively, right ovarian mass of 6 – 8 cm size was seen attached to uterus, pelvic peritoneum, caecum and appendix with omental adhesions. Frozen section was performed which did not show any malignancy. The gross specimen received comprises of 8.5x5.5x3 cm. ovarian mass with smooth capsulated external surface. Cut surface showed multiple cysts filled with dirty fluid. The cyst contents sent for culture did not show any micro organism growth. Histopathological examination revealed dense cellular infiltrate comprising of foamy histiocytes, neutrophils, lymphocytes, plasma cells and eosinophils (Fig 1). No tumor was identified even after extensive sampling. The histopathological diagnosis of Xanthogranulomatous oophoritis was rendered.

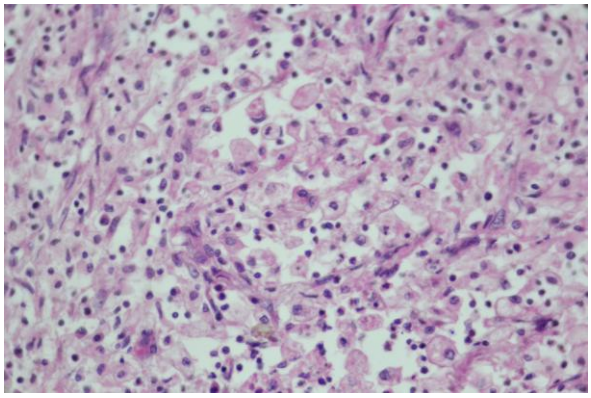

Fig 1: Effaced ovarian parenchyma with dense chronic inflammatory cell infiltrate and foamy histiocytes (H&E stain, 600X)

Case 2: A 31 year old married women presented with lower abdomen pain, burning during micturition, difficulty in passing stools and fever. Her medical history was unremarkable and on examination no lump was felt on pelvic examination. Her hematological findings showed slightly raised TLC (10.74 $x10^9$/L) with neutrophilia, urine examination showed 20 – 25 pus cells/ hpf. Colonoscopy showed internal haemorrhoids grade II. Pleural fluid examination did not show any abnormal cells. Urine culture was performed did not show any growth. Patient underwent exploratory laprotomy with total abdominal hysterectomy and bilateral salpingo-ophrectomy, sigmoid colectomy and supracolic omentectomy. Per operatively, abscess was noted between left ovarian mass and sigmoid colon. Gross examination showed large left tubo ovarian mass measuring 4.5x4x3 cm. with multiloculated cystic cavity filled with serous fluid. The mass showed extensive adhesions to left tube and sigmoid colon. Cervix showed a 1.2x0.5x0.3 cm firm grey white area. Rest of the uterus was unremarkable. Microscopic examination of the ovarian mass revealed ovarian parenchyma showing cyst wall (Fig 2) infiltrated by dense mixed inflammatory cell infiltrate (Fig 3) comprising of histiocytes, plasma cells, neutrophils, fibroblasts, and lymphocytes. Micro sections from cervix revealed well differentiated squamous cell

carcinoma. Left tube showed chronic salpingitis and serositis of sigmoid colon was noted. The histopathological diagnosis of xanthogranulomatous oophoritis was made.

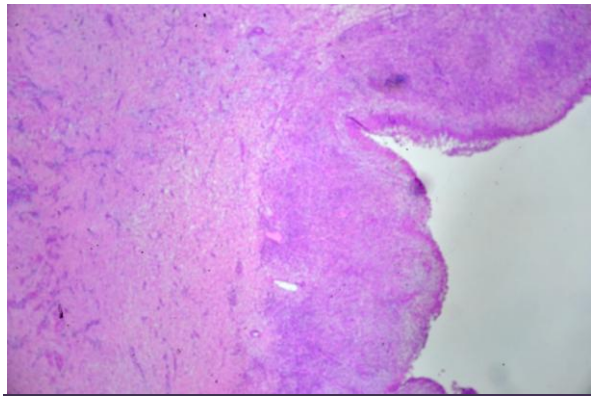

Fig 2: Effaced ovarian parenchyma with cyst wall lined by dense chronic inflammatory cell infiltrate (H&E stain,100X)

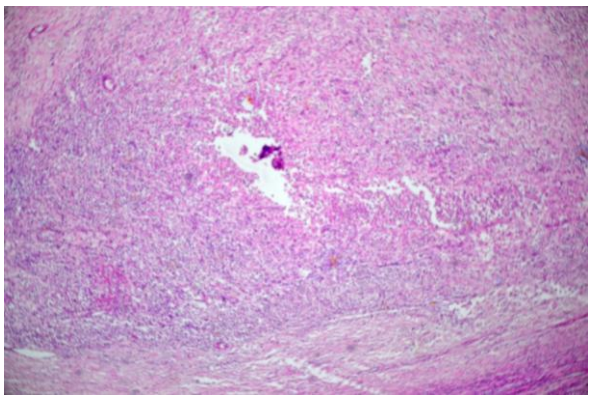

Fig 3: Dense chronic inflammatory cell infiltrate and foamy histiocytes (H&E stain, 400X)

Discussion

Kunakemakorn was first to describe xanthogranulomatous inflammation of the serosa of uterus, left fallopian tube and ovary in his report of inflammatory pseudo-tumor in the pelvis in 1976. The pathogenesis of xanthogranulomatous inflammation remains unclear. The proposed causes of foam cell production are infection, ineffective antibiotic therapy, abnormality in lipid metabolism, endometriosis and ineffective clearance of bacteria by phagocytes [3,6,7,]. Bacterias like B. fragilis, E. coli, S. aureus and S. typhi can be considered in the pathogenesis of this form oophoritis. [1,4].

The average age of patient of xanthogranulomatous oophoritis is 31 years [3,4] as in our cases. The clinical presentations include fever, abdominal mass, pain in the abdomen, menorrhagia, anemia and anorexia [1,4] as observed in our cases. Clinically and radiologically xanthogranulomatous inflammation may mimic ovarian tumor [1].

Contrast enhanced CT shows complex solid-cystic lesions with thick enhancing walls and variably enhancing solid intramural nodules [4]. MR imaging findings of xanthogranulomatous oophoritis are multiple intramural nodules in a thickened wall with high signal intensity on T2 – weighted images and low signal intensity on T1-weighted images [3].Grossly in xanthogranulomatous inflammation, the affected ovary can be replaced by a well circumscribed, solid, yellowish, lobulated mass but can also present with cystic lesion at times [1, 6] as in our cases. Microscopically, the lesion is characterized by lipid laden histiocytes. The proliferation of foamy histiocytes and fibrous obliteration of the chronic inflammatory process are responsible for the pseudotumoral appearance on gross examination [4].

Xanthogranulomatous inflammation often poses diagnostic challenge for pathologists because of its rarity. Both non neoplastic and neoplastic conditions are fielded in the differential diagnosis. Because of the presence of foamy histiocytes, malakoplakia is also to be considered in the differential diagnosis [8]. According to Wather, common pathogenetic pathway is noted for both malakoplakia and xanthogranulomatous inflammation. In malakoplakia, the cytoplasmic concenteric calcific bodies (Michaelis-gutmann bodies) are found (absent in our case) [1,4]. If the lesion shows mainly sheets of lymphocytes, it may be misdiagnosed as secondary lymphoma or leukemia. If the lymphocytes are scattered diffusely and foam cells are scare, there is a possibility of misdiagnosis of the condition as malignant small cell tumor. If there are small amount of obvious fibrous and foam cells, a diagnosis of sclerosing stromal tumor can be made.

Frozen section is very helpful diagnostic modality in such cases where xanthogranulomatous inflammation is mimicking like a tumor. Immunohistochemistry stains are helpful in demonstrating the histiocytes (CD68) thus consolidating the diagnosis of xanthogranulomatous oophoritis. The treatment of choice for xanthogranulomatous oophoritis is oophrectomy. [2,6] Awareness of this inflammatory lesion can prevent radical surgery and also misdiagnosis as a malignancy. [1]

Conclusion

Xanthogranulomatous inflammation of the female genital tract is rare and ovarian involvement as such is further rarer. It commonly mimics ovarian neoplasm leading to diagnostic dilemma for clinicians as well as for pathologist. However peroperative frozen section and histopathological examination can enlighten the inflammatory pathology and help in proper management.

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Competing Interests

None declared.

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