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Xanthogranulomatous Salpingo-Oophoritis: Case Report of a Rare Entity

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Abstract **Case Report**

Xanthogranulomatous inflammation is a chronic non-neoplastic condition where affected organ is replaced by Xanthoma cells alongwith other chronic inflammatory cells. Female genital organs are very rarely involved by the disease. Such inflammation occurring in a thirty-six year old patient is reported here. The presence of intrauterine contraceptive device (IUCD) probably contributed to the development of this disease in her case. She was successfully treated by conservative surgery. The need to keep patients carrying IUCD under close surveillance to prevent development of this disease in such patients is emphasized in this report.

Keywords: Xanthogranulomatous inflammation, Salpingitis, Oophoritis.

INTRODUCTION

Xanthogranulomatous inflammation is a chronic non-neoplastic condition in which the affected organ is replaced by lipid-filled macrophages, plasma cells, lymphocytes and neutrophils [1]. Usual organs involved are kidneys, urinary bladder, stomach, testes, bones, gall bladder and epididymis [2]. Involvement of female genital organs is very rare and endometrium is mainly involved [3]. Only a few cases of involvement of ovaries and fallopian tubes have been reported till date [4, 5]. Exact etiology of this type of inflammation is not known although multiple predisposing factors such as pelvic inflammatory disease, IUCD use, leiomyoma uterus, endometriosis and inappropriate antibiotic intake are attributed to its development [6]. A case of xanthogranulomatous salpingo-oophoritis in a premenopausal multiparous woman with IUCD in place and presenting with fever and lower abdominal pain is being reported here.

CASE REPORT

A 36 year old premenopausal woman presented to the Gynaecological OPD of a tertiary care Hospital in July 2017 with complaints of fever and pain in left lower abdomen. She had intrauterine Copper-T in place. She was clinically examined and diagnosed to be having pelvic inflammatory disease with left tubo-ovarian mass. Apart from positive Widal test, predominantly neutrophilic leucocytosis and raised ESR, other investigations as to the cause of her fever were within normal limits. Abdominal ultrasound raised suspicion of cystadenoma of left ovary. IUCD was seen in place. Magnetic Resonance Imaging of abdomen raised possibility of left tubo-ovarian abscess with hemorrhagic cyst within left ovary.

Her Copper-T was removed and she was started on antibiotics. As her fever did not respond to antibiotics and tubo-ovarian mass showed no evidence of shrinking, laparotomy and left salpingo-oophorectomy was done. Peroperatively uterus was of normal size and right adnexa were normal. Examination of left adnexa revealed pyosalpinx from which 100 cc of pus was drained and sent for culture (positive for Staph. aureus). An inflamed left ovarian mass 9 x 10 cm was present adherent to surrounding gut and to round ligament. On gross histopathological examination of the operated specimen (Fig. 1), a soft grayish-white ovarian mass with focal congested areas was seen. Cut section showed multiple dark brown hemorrhagic areas. Cut section of fallopian tube showed greyish-white material. On microscopic examination (Fig. 2 & 3), multiple sections from the ovary showed presence of diffuse and

dense infiltration of ovarian stroma by foamy histiocytes, lymphocytes, plasma cells and neutrophils. Also seen were congested and thrombosed blood vessels, areas of fibrosis and hyalinised stroma. AFS and PAS staining were negative. Sections studied from fallopian tube also showed above described lesion. A histopathological diagnosis of Xanthogranulomatous Salpingo-oophoritis was made. Because a firm histopathological diagnosis could be arrived at, immunohistochemical staining for CD68, a histiocytic marker in foamy histiocytes, CD3, a T cell marker, and CD20, a B cell marker in the background lymphocytes, was not used.

The patient recovered well after surgery and was discharged on seventh post-operative day. She became afebrile and had no pain abdomen.

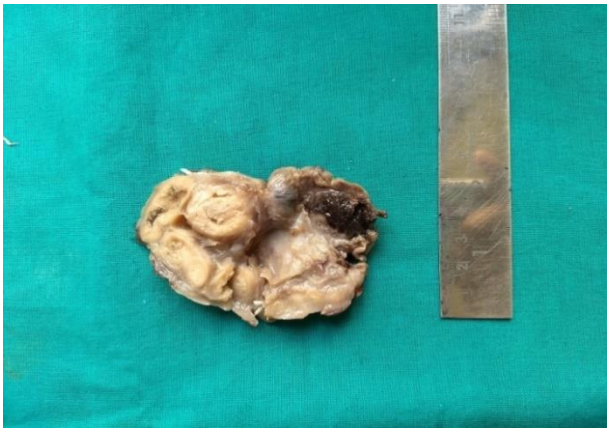

Fig-1: Gross examination shows gray white mass replacing the tubes and ovaries

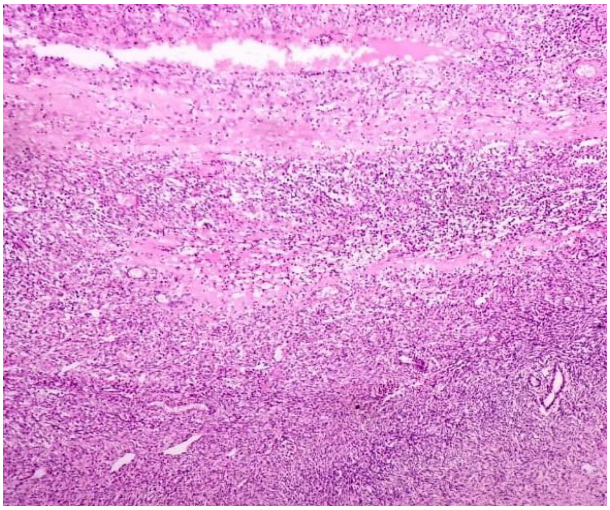

Fig-2: H&E (10X) shows diffuse and dense infiltration of ovarian stroma by foamy histiocytes, lymphocytes, plasma cells and neutrophils

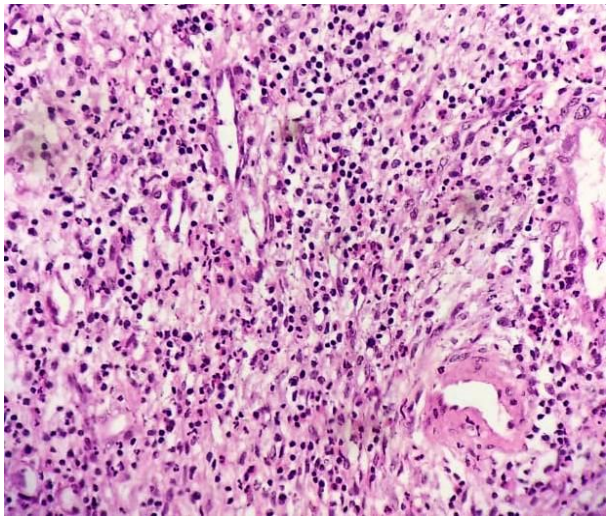

Fig-3: H&E (40X) shows diffuse and dense infiltration of ovarian stroma by foamy histiocytes, lymphocytes, plasma cells and neutrophils

DISCUSSION

Xanthogranulomatous inflammation of uterus, fallopian tube and ovary was first described by Kunakemakorn[7] in 1976. The average age reported is 38.5 years [6] though our patient was 36 years old. The clinical presentations include anemia, anorexia, fever, irregular colporrhagia and hypogastric pain. Gynaecological examination reveals tender adnexal mass [6]. Longstanding pelvic inflammatory disease refractory to antibiotic treatment is often reported by patient. The patients, like in this case, often have IUCD in place. It is thought that the colonization of the IUCD in the endometrial cavity is followed by bacterial shedding through fallopian tube lumen. At time of ovulation, the ruptured ovarian surface gets exposed to these bacteria leading to infection of corpus luteum and production of ovarian abscess [8]. The responsible bacteria that have been associated with xanthogranulomatous inflammation include Escherichia coli, Salmonella typhi, Staphylococcus aureus etc[9]. Xanthogranulomatous inflammation in female genital tract mainly affects one side [6]. Elevated ESR and leucocytosis are often found on laboratory tests as in this case. Ultrasound, CT and MR scans demonstrate a well-defined solid mass which may mimic malignancy or abscess, like in this case [6]. Histopathological examination of affected organs shows disorganization and infiltration with focal or sheets of foam cells (called Xanthoma cells) admixed with chronic inflammatory cells such as lymphocytes, plasma cells, occasional neutrophils with or without multinucleated or Touton giant cells [6]. Since it is a chronic inflammatory process leading to tissue necrosis, the continuous release of cholesterol and other lipids from the dead cells, phagocytosed by macrophages, leads to xanthomatous process [10]. Among conditions which may be confused with xanthogranulomatous inflammation on microscopy include secondary lymphoma or leukemia if the lesion is mainly focal

scattered lymphocytes. If the lymphocytes are diffusely scattered and foam cells are few, a diagnosis of malignant small cell tumour with stromal luteinization may be put forward. In presence of small amount of obvious fibrosis and foam cells, a diagnosis of sclerosing stromal tumour may be entertained [6]. Wather considered malakoplakia and xanthogranulomatous inflammation as identical chronic inflammatory diseases. However, in malakoplakia, the cytoplasmic concentric calcific bodies (Michaelis – Gutmann bodies) are seen [11], but these were not seen in our patient.

Xanthogranulomatous inflammation is also reported to be lethal by causing systemic inflammation. Hence, aggressive treatment in form of surgery is recommended but surgery need not be as radical as it would necessarily have been had it been a malignant process [12, 13].

CONCLUSION

As xanthogranulomatous inflammation is a locally destructive form of chronic inflammation, prevention and early diagnosis is the key to prevent extensive damage of the involved organs like the female genital tract. It has been suggested that patients with pelvic inflammatory disease, endometriosis and those carrying IUCD should be kept under close follow-up to prevent occurrence of this disease [5].

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