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CODEN: IJBABN **Case Report**

A rare ovarian mass with pregnancy: A Case Report

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Abstract

Xanthogranulomatous inflammation (XGI) is an uncommon, non-neoplastic, chronic process in which the affected organ is destroyed by massive cellular infiltration of foamy histiocytes admixed with multinucleated giant cells, plasma cells, fibroblasts, neutrophils. The etiology of this entity is unknown. It is more commonly seen in the kidney, Intestines and gall bladder. Very few cases of Xanthogranulomatous Oophoritis (XO) are relatively rare and so far no case exists of this condition affecting the ovary during pregnancy.

We report here a 30-year-old Primigravida @ 35 wks PoG with twin gestation, who presented with a large ovarian mass with suspicion of infective pathology on clinico-radiological findings and diagnosis of xanthogranulomatous oophoritis, which was established on histopathology.

Clinically and radiologically, xanthogranulomatous oophritis mimics tumor of the ovary thereby making it an important entity. A thorough histopathological examination can aid in confirming the diagnosis.

Keywords: Xantogranulomatous Oophorotis, Foamy Histiocytes, Chronic inflammation.

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1. Introduction

Xanthogranulomatous inflammation is an uncommon form of chronic inflammatory process that is destructive to normal histology [1]. It is characterized by replacement of normal tissues of the affected organ by lipid containing macrophages with an admixture of lymphocytes, plasma cells and neutrophils.

The most affected organs are the kidney and gallbladder, followed by anorectal area, bone, stomach, bladder and testis [2]. In female genital tract, it more commonly affects the endometrium but effect on vagina, cervix, fallopian tube, and ovary can also be seen [3]. Kunakemakorn was the first to report inflammatory pseudo-tumour in the pelvis in 1976[4]. The clinical manifestations are pelvic masses, which can easily be misdiagnosed as ovarian tumors or tuberculosis. The clinical manifestations, radiological and macroscopic features of this condition are similar to ovarian tumors.

Very few cases are reported in literature of this condition and the current case is therefore reported in view of its rarity [5].

2. Case Report

Thirty years old lady, a known case of bilateral endometrioma conceived twins after IVF. She initially presented to our centre in second trimester at approximately 14 weeks with acute onset of pain abdomen with fever. She underwent emergency laparotomy where the infected ovarian abscess was drained of the pus. There were dense adhesions between the ovary and bowels and no further dissection or adhesolysis was attempted. She was allowed to continue the pregnancy.

Subsequently she continued to have brief episodes of pain abdomen which were managed conservatively. She had a twin pregnancy that continued uneventfully till she

again presented at 35 weeks with complaints of pain abdomen and fever. Clinically she was found to have a tender mass alongside the gravid uterus extending from the Pouch of Douglas to the lumbar region. Imaging including MRI revealed a large mass, 15x13x19 cms, occupying the entire POD and extending to the lumbar region. The fat planes were preserved. There was no fetal compromise. In view of the distress caused to the mother and the fetuses being uncompromised, she was taken up for Elective CS and exploratory laparotomy.

2.1 On evaluation:

Antenatal MRI -Heterogeneous mass in POD (15 *13*19 cms). Extending superiorly upto L3- L4, Inferiorly reaching upto coccyx, anteriorly abutting posterior surface of uterus and posteriorly abutting lumbosacral vertebrae from bifurcation of aorta to coccyx. B/l ureters were displaced posteriorly and laterally.

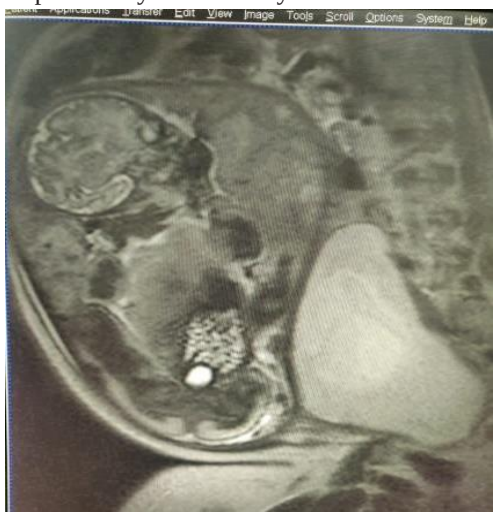


Fig 1A: MRI showing sagittal view of mass occupying whole of POD

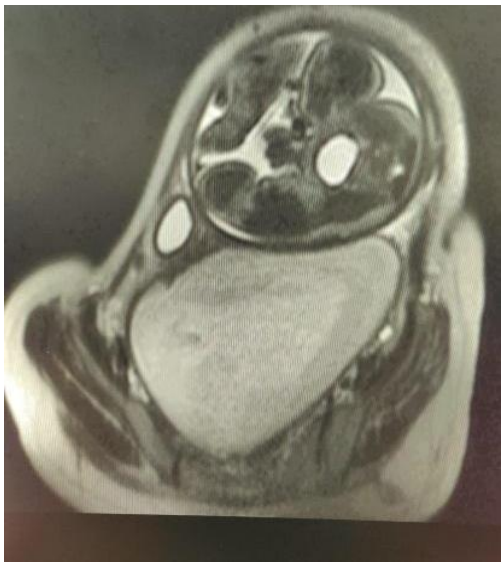
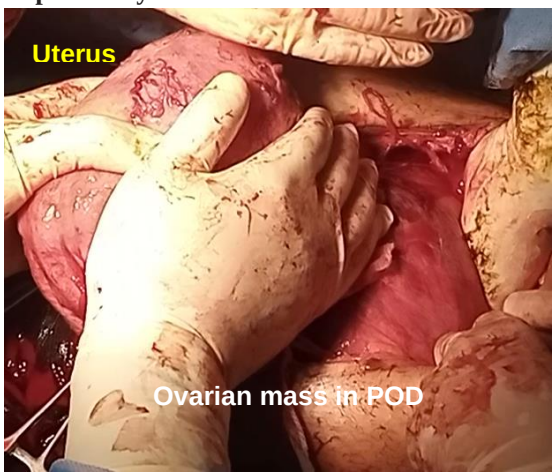


Fig 1B: MRI in transverse view showing the posterior extent of mass

2.2 Tumour markers:

Ca 125- 26.8 IU/L, HE 4 – 172.50 ng/ml
Ca 19.9 - <2 IU/L, CEA - < 0.5 IU/L

Intra operatively:



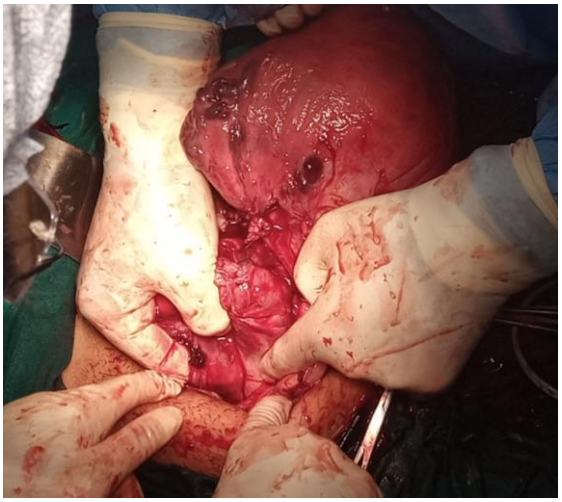


Fig 2: Huge Rt ovarian mass occupying POD, adherent to bilateral pelvic wall, uterus and rectosigmoid

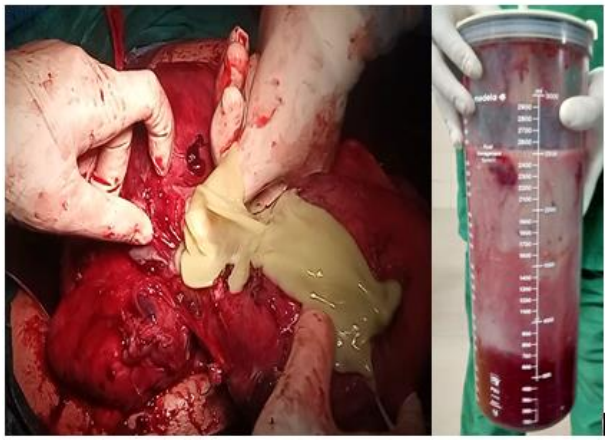


Fig 3: 2.5 Litres of pus drained from the mass

2.3 Microbiological examination of Pus revealed:

Gram stain: *Klebseilla pneumoniae*

Sensitive to Gentamicin and colistin

ZN stain: No AFB seen, PAS stain: No fungi seen

LPA (line probe assay): No atypical mycobacteria

Gene Xpert: No MTB

She was treated with broad spectrum antibiotics for 7 days and her Post – op period was comfortable.

Histopathological examination revealed the diagnosis of xanthogranulomatous oophoritis, which is a relatively rare condition and so was reported independently by 2 pathologists. Further on IHC -CD 68 was positive.

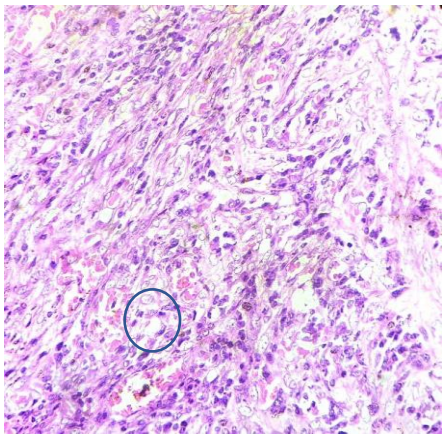


Fig 4 A: Multiple foamy histiocytes: Pathognomonic of Xanthogranulomatous oophoritis

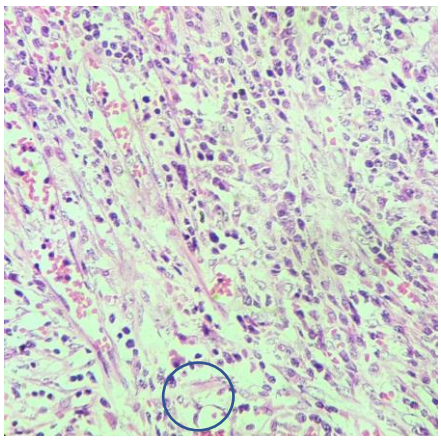


Fig 4B: Plasma cell infiltration

3. Discussion

Xanthogranulomatous inflammation is characterized by destruction of the tissues of the organ involved and they are replaced by chronic inflammatory cells such as lymphocytes, plasma cells, neutrophils and foamy histiocytes.

Xanthogranulomatous inflammation of the female genital tract is rare and when it affects the female genital tract, affects the endometrium [10]. Fallopian tubes or ovaries focally or entirely, which clinically forms mass-like lesion in the pelvic cavity and can involve the surrounding tissues is rare.

It mostly affects women of reproductive age group. However, the youngest documented case report of xanthogranulomatous oophoritis is of a 2-year-old girl [6]. Another girl who was just 18 years of age was also reported to have xanthogranulomatous inflammation of the ovaries [7].

The exact pathogenesis of xanthogranulomatous oophoritis is still unknown and unclear. Many theories have been postulated. They are various associated conditions like infection, endometriosis, intrauterine contraceptive device, inborn errors of lipid metabolism, and drug induced. Infection is the most consistent of associations and

Microorganisms such as *Escherichia coli*, *Proteus* spp., *Staphylococcus aureus*, *Bacteriodes fragilis*, *Salmonella typhi*, *Actinomyces*, *Streptococcus (Enterococcus) faecalis*, viridans streptococci, and group B streptococci are implicated in this condition [8].

Punia et al described xanthogranulomatous oophoritis and salpingitis as late sequelae of inadequately treated pelvic inflammatory disease caused by *Staphylococcus* spp [9]. But our patient had grown Klebseilla pnemoniae, which is very rare and likely to be the first in literature.

Shukla et al described an association of this entity with primary infertility and endometriosis [10]. Our patient too had primary infertility and endometriosis.

Patients usually present with a history of inadequately treated pelvic inflammatory disease. [11]

The diagnostic dilemmas presented by such masses are mostly because they mimic a tumour that may be benign or malignant. The clinico-radiological findings are very similar in both. The literature shows several cases that were misdiagnosed as ovarian cancers [12]. Hence, clinicians, especially Gynecologists need to be aware of this entity and should be able to differentiate it from endometrial, ovarian, or tubal malignancy.

The extent of the masses reported in literature searches extend up to 10cm and inflammation when extends to the neighbouring organs; pelvic structures and peritoneal including bowel and abdominal walls. The dimensions of the mass in our patient were much larger, being 19 x 13 x 8 cms. The masses are usually solid, fragile, and accompanied by areas of haemorrhage, necrosis, and cystic degeneration due to liquefactive necrosis [5]. The cross section is solid with areas of haemorrhage, necrosis and purulent collection, as was seen in our case too, where 2 litres of purulent fluid was found.

Histologically, the normal ovarian structure is destroyed and replaced by chronic inflammatory cells

where sheets of foam cells, foamy histiocytes, ill formed granulomas, multinucleated giant cells, inflammatory cells and fibroplasia. Neo vascular proliferation is also seen, which gives the gross appearance of a tumor. [13]

The presence of foamy histiocytes adds an additional differential diagnosis to this condition: Malakoplakia. It is thought to represent an inflammatory process in which macrophages do not have the adequate ability to kill the bacteria after phagocytosis. Malakoplakia characteristically shows the cytoplasmic concentric calcific bodies known as Michaelis-Gutmann bodies which are absent in xanthogranulomatous inflammation. This condition is mainly seen in the urinary system.

In our country, important differential diagnoses include tuberculosis and fungal infections, which are ruled out by special stains and culture studies. Accordingly in this patient, this patient, ZN stain and PAS stain were negative, thereby ruling out tubercular and fungal infection.

Due to the presence of lymphocytes, secondary lymphoma/leukemia can also be considered as differentials. But in lymphoma/leukemia there is paucity of foam cells, Immunohistochemistry can be helpful to establish the diagnosis with CD68 (foam cells positive), CD3 (T lymphocyte marker), CD20 (B-lymphocyte marker), and κ and λ (both positive in polyclonal B-lymphocytes).

An extensive literature search revealed very few cases reported in pregnancy. This makes our case extremely rare. [12]

4. Conclusion

It is evident that Xanthogranulomatous oophoritis is a very rare condition, which can often be a diagnostic challenge in clinical practice as it mimics conditions like malignancy. It is important for clinicians to be aware of this entity and be vigilant to prevent a misdiagnosis of malignancy. The cornerstone of the diagnosis in awareness and a meticulous history taking and clinical examination, aided by relevant investigations and confirmed by diligent histopathological examination.

Conflict of interest:

The authors declare that there was no conflict of interest.

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