Case Report

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A Rare Case of Xanthogranulomatous Salpingo-oophoritis

Ananya Das[1], Megha Goyal[2], Ahanthem Santa Singh[3], Biswajit Dey[4], Kothuni Christina Chakre[5], Namitha Gowda[6]

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ABSTRACT

Background: Xanthogranulomatous salpingo-oophoritis is an infrequent and challenging diagnosis of the female genital tract. It involves the destruction of the fallopian tube and ovarian tissue by infiltrating inflammatory cells comprising lipid laden macrophages, lymphocytes, plasma cells, and multinucleated giant cells. While more commonly found in gall bladder and kidney, its occurence in female genital tract is rare. This is a case of Xanthogranulomatous salphingo-oophoritis in a 48 year woman who presented with chronic lower abdomen pain and frequent heavy menses for two months. On per abdomen examination a non tender 24 weeks size mass was felt and on vaginal examination the same mass was appreciated, seperate from the uterus. PAP was reported NILM. Ultrasound revealed uterine adenomyoma with bilateral endometrioma of ovaries and associated left hydrosalphinx. Patient underwent hysterectomy with bilateral salphingo-oopherectomy. Histopathological examination revealed uterine adenomyosis, chronic cervicitis with xanthogranulomatous salpingo-oophoritis. The patient recovered well and doing good on follow-up.

Conclusion: Xanthogranulomatous salpingo-oophoritis, though rare is a significant entity because clinically it can mimic chronic pelvic inflammatory disease or endometrioma and even in some cases malignancy. Even clinical examination and imaging might not help much. Therefore, knowledge and awareness about the condition with the help of histopathology can clinch the diagnosis and also prevent radical cancer surgery.

Keywords: Xanthogranulomatous; Lipid-laden; Salpingo-oophoritis; Histopathological; Hysterectomy.

Author's Affiliation: [1]Additional Professor, [2]Post Graduate 2nd year Student, [3]Senior Professor and HOD, [5]Senior Resident, [6]Post Graduate Student, 3rd year, Department of Obstetrics and Gynecology, [4]Associate professor, Department of Pathology, North Eastern Indira Gandhi Regional Institute of Health & Medical Sciences, Shillong 783018, Meghalaya, India.

Corresponding Author: Ananya Das, [1]Department of Obstetrics & Gynecology, North Eastern Indira Gandhi Regional Institute of Health & Medical Sciences, Shillong 783018, Meghalaya, India.

E-mail: mailmedrananyadas@rediffmail.com

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INTRODUCTION

Xanthogranulomatous salphingo-oophoritis is an infrequent and challenging diagnosis of the female genital tract. It involves the destruction of the fallopian tube and ovarian tissue by infiltrating inflammatory cells comprising lipid laden macrophages, lymphocytes, plasma cells, and multinucleated giant cells.[1] While more commonly found in gall bladder and kidney, its occurence in female genital tract is rare. Xanthogranulomatous salpingo-oophoritis has been previously reported in the literature under various names,

including xanthogranulomatous oophoritis, xanthogranulomatous inflammation of the ovaryand fallopian tube, and lipid cell granuloma of the ovary. In 1968 Roth published the first article describing a case of xanthogranulomatous oophoritis and since then, there have been almost 100 cases documented in the literature. Xanthogranulomatous oophoritis has an uncertain specific aetiology, although it's been postulated to result from chronic inflammation, infection, or autoimmune disorders.[2] However due to its locally destructive nature and mass forming capacity as a result of adhesions, this type of inflammation may sometimes mimic malignancy both clinically and radiologically. Therefore, awareness of this entity is of paramount importance in proper management of such patients.[3]

CASE REPORT

48 year woman P4L4 presented with chronic lower abdomen pain and frequent heavy menses for two months. There was no history of any chronic illness like tuberculosis in the past. General Examination: No Pallor, Icterus, Cyanosis, Clubbing, Lymphadenopathy, Edemanoted. On per abdomen examination, a non tender 24 weeks size mass was feltand on vaginal examination, same mass was appreciated, seperate from the uterus. PAP Smear was reported Negative for intra epitheliallesion or malignancy (NILM). Ultrasound revealed Bilateral mild Hydroureteronephrosis with Uterine adenomyoma with bilateral adnexal cystic lesion suggestive of endometrioma (Right: 13*10 cm) (Left: 6*4 cm) with left hydrosalphinx

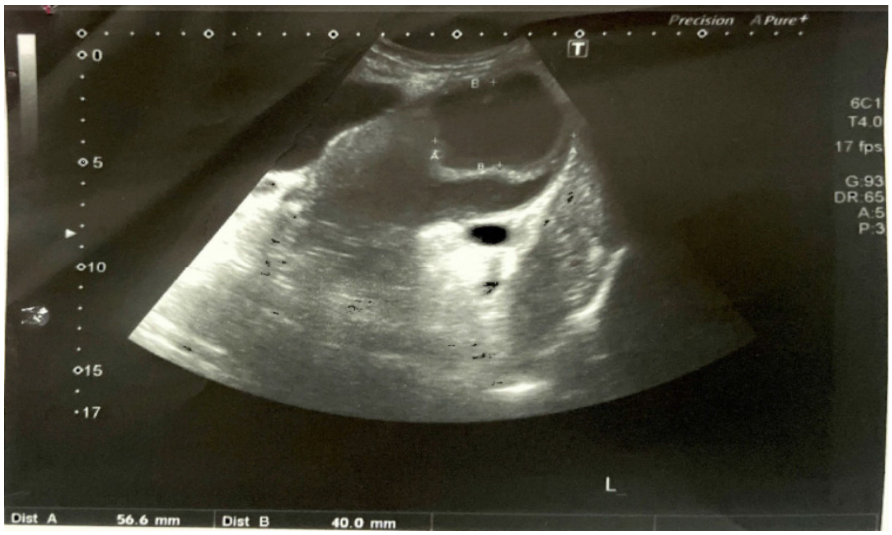

Fig. 1: Left sided Endometrioma (6*4 cm)

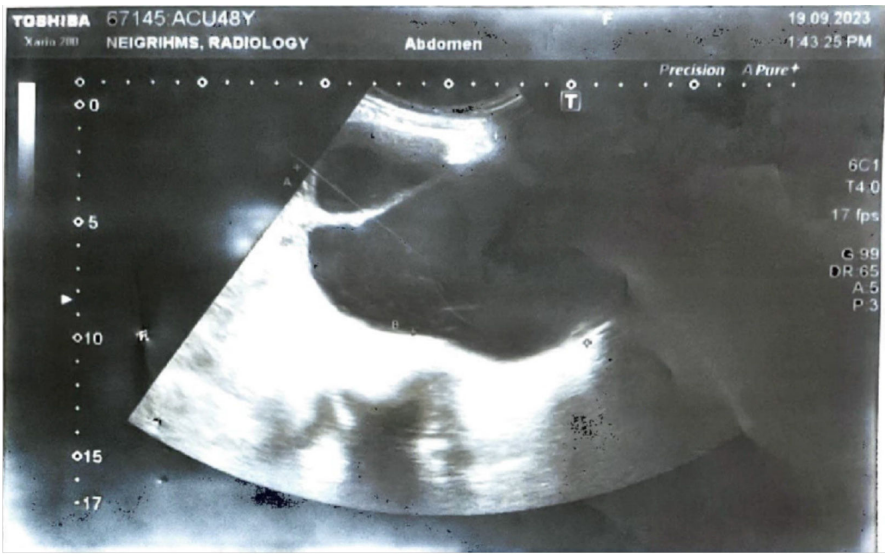

Fig. 2: Right sided endometrioma (13*10 cm)

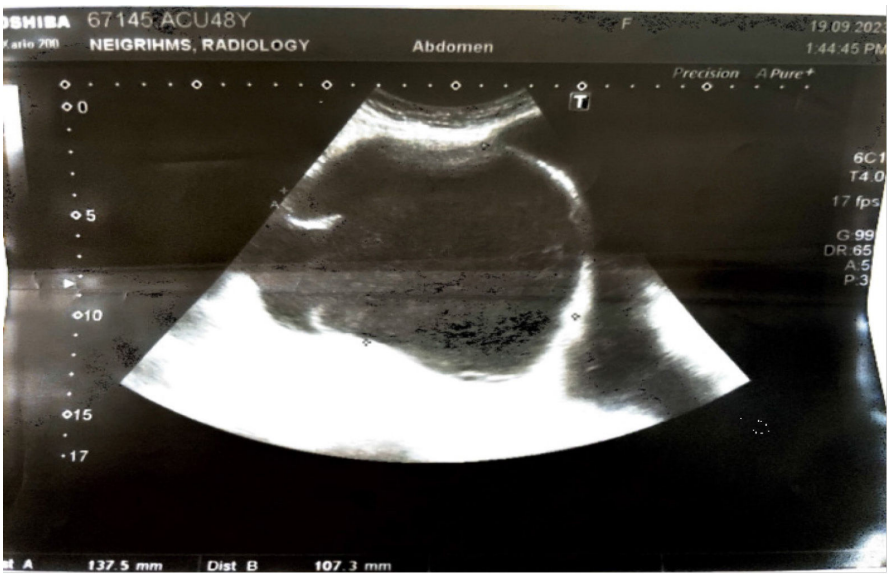

Fig. 3: Left sided Hydrosalphinx (11*8.4 cm)

(11*8.4 cm).

Patient underwent hysterectomy with bilateral salphingo-oopherectomy with bilateral ureteric stenting.

Intra-operative: Uterine size was found to be 10-12 weeks with Bilateral tubo-ovarian mass Right (~15*12 cm) >Left (6*7 cm). Bilateral ovarian mass was found densely adhered to bowel posteriorly.

Frozen section revealed features suggestive of benign cyst.

Final Histopathological examination revealed uterine adenomyosis, chronic cervicitis with xanthogranulomatous salphingo-oophoritis.

Fig. 4 & 5: Low and high power histopathology images showing sheets of foamy histiocytes admixed with lymphocytes and occasional neutrophils (H &E, 40x and 400x).

The patient recovered well and doing good on follow-up.

DISCUSSION

Xanthogranulomatous inflammation involving the female genital tract is an uncommon and distinct kind of persistent inflammation with tissue destruction in the affected organs[4] However, it is less common in the female genital tract with only a handful of cases been reported. This has been found in patients aged 21-75 years with a mean age of 45 years.[5]

The pathogenesis of xanthogranulomatous oophoritis is unclear and many theories that are of etiopathogenesis have been postulated, such

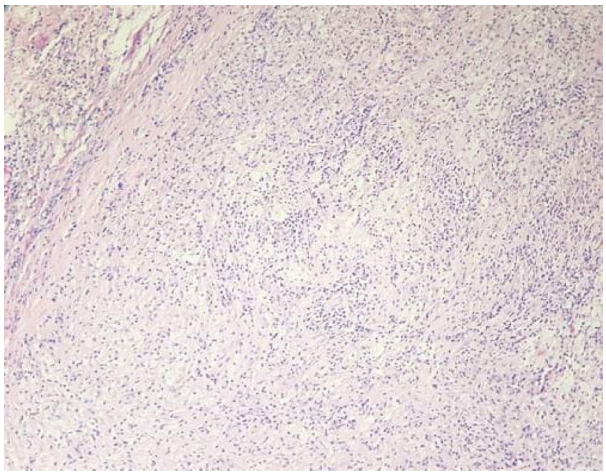

Fig. 4

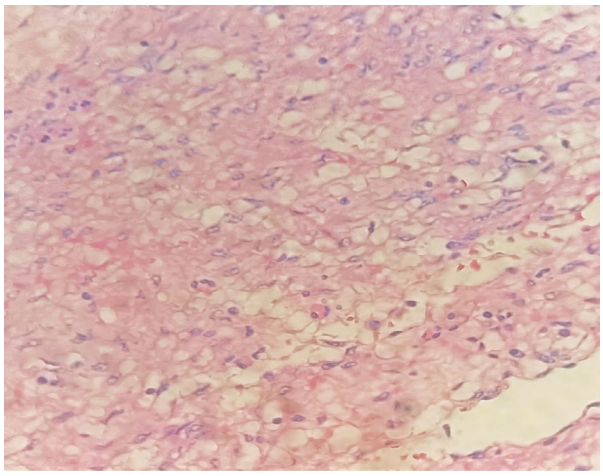

Fig. 5

as theory of infection, endometriosis, intrauterine contraceptive device, inborn errors of lipid metabolism, and drug induced. Amongst these theories, the most accepted theory is of infection, which is supported by clinical evidence of infection and growth of bacteria such as *Escherichia coli*, *Bacteroides fragilis*, and *Proteus vulgaris* from the affected tissue by culture.[6] Another explanation for this phenomenon is tissue necrosis, which is brought on by a prolonged infection and results in the ongoing release of lipids and cholesterol from the dead cells. The xanthomatous process begins as a result of macrophages phagocytosing these biological components.[8]

Radiological findings of xanthogranulomatous oophoritis may simulate endometriosis or 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, there by mimicking malignancy.[7] Microscopically, there is infiltration of sheets of foamy cells admixed with mixture of inflammatory cells such as lymphocytes, plasma cells, neutrophils with multinucleated giant cells.[1] 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 Inefficient or inappropriate antibiotics applied in the early phase of infection that resulted in ineffective control of bacterial multiplication or Presence of a lipid metabolic disorder that induces hyperlipidemia and the foam cells are formed when the lipid deposited is phagocytosed by phagocytes.[4]

Differential diagnosis of xanthogranulomatous oophoritis includes infections like tuberculosis, fungal infections which can be ruled out by culture and special stains for the causative organisms. Malakoplakia is also one of the differential diagnoses.[9] In malakoplakia, the basophilic cytoplasmic concentric calcific bodies within histiocytes (Michaelis-Gutmann bodies) are found which were absent xanthogranulomatous inflammation ruling out this condition.[9] Granulomatous salpingo-oophoritis in surgical pathology practice in developing countries may be seen in many other conditions most common being tuberculosis. Others include a foreign body reaction to suture material introduced at a previous operative procedure, associated Crohn's disease, previous diathermy, a necrotizing reaction following previous surgery, endometriosis and bacterial tubo-ovarian abscess. In few cases no cause could be attributable for the granulomatous inflammation and small cortical granulomas in the ovary are seen called as idiopathic granulomas.[10] Antibiotic therapy has been attempted, but it has not succeeded in reducing ovarian mass.[11]

As done in our case, surgery is the treatment of choice. But an Awareness of this inflammatory lesion among the clinicians, radiologists and pathologists may not only prevent overdiagnosis and extensive surgeries for the patients but may also reduce morbidity giving a better prognosis to these patients

CONCLUSION

Xanthogranulomatous salphingo-oophoritis, though rare is a significant entity because clinically it can mimic chronic pelvic inflammatory disease or endometrioma and even in some cases malignancy. Even clinical examination and imaging might not help much. The final confirmation of the diagnosis can only be made after histopathological examination. It must be kept as a differential diagnosis specially in cases with complex tubo-ovarian masses.[6]

Therefore, knowledge and awareness about the condition with the help of histopathology can clinch the diagnosis and also prevent radical cancer surgery.

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