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A rare case report of xanthogranulomatous oophoritis with serous cystadenoma in pregnancy

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Abstract
Xanthogranulomatous oophoritis is a rare non neoplastic chronic inflammatory process of ovary which causes functional failure of the organ due to gross structural destruction. We report a case of 32 year old female gravid 2 para 0 with 1 history of ectopic pregnancy who was diagnosed with a cystic mass lesion of right ovary by USG and MRI at 20 weeks of gestation. After an uneventful antenatal period, she delivered a live healthy male baby at 39 completed weeks of gestation by caesarean section. Right sided oophorectomy was done and microscopic examination of the specimen showed serous cystadenoma with ovarian stroma infiltration with foamy histiocytes, inflammatory cells and multinucleated giant cells. This case is reported because no case as such is mentioned in literature.

Keywords: xanthogranulomatous oophoritis, serous cystadenoma, pregnancy

1. Introduction
Xanthogranulomatous oophoritis is a non-neoplastic chronic inflammatory process in which the normal anatomy of affected organ is destroyed and is replaced by massive cellular infiltration of foamy histiocytes admixed with multinucleated giant cells, plasma cells, fibroblasts, neutrophils, and foci of necrosis [1]. Xanthogranulomatous inflammation of ovary causes functional failure of the organ due to gross structural destruction. It is an uncommon process, most commonly affects kidney and gall bladder, followed by anorectal area, bone, stomach, and testis [1]. Only a few cases involving the ovary and fallopian tube have been reported [2, 3] Till date, 46 cases of salpingo oophoritis were published in literature out of which 32 case reports are described in India literature [4] After extensive search of literature, we found no case report of serous cystadenoma with xanthogranulomatous oophoritis in pregnancy. So here we report a case of serous cystadenoma with xanthogranulomatous oophoritis in a pregnant woman who also had a past history of ectopic pregnancy affecting the same sided fallopian tube as that of the ovarian lump.

2. Case report
A 32 year old female who was gravid 2 para 0 with 1 history of ectopic pregnancy presented at 20 weeks of gestation for antenatal checkup. She was advised an obstetrical Ultrasonography as a part of routine antenatal investigation. Her USG report showed a live intrauterine gestation of 19 weeks 5days +/- 1 week, Placenta was lying posteriorly in upper and mid body region. Liquor was adequate and FHR was 138 beats/minute. There was also a hypoechoic mass lesion in right adnexa of ovarian origin measuring 6.9x5.1 cm^2. Colour Doppler study showed increased vascularity. However the patient had no complains as such. MRI whole abdomen showed a gravid uterus along with a 6x5.5 cm^2 well circumscribed complex cystic lesion in the right ovary at posterior cul-de-sac. It was hypo intense on T1W images and heterogeneously hyperintense on T2W images. The capsule was intact. Serum CA125 was 17 U/ml, Serum CEA was 0.87ng/ml and Serum AFP was 62.10ng/ml (which was within normal limits as per her gestational age). She had regular gynaecological follow up before conceiving this time and her pelvic ultrasonography done 2 months before conceiving showed both ovaries normal (Right ovary 38x20mm, Left Ovary 27x13mm).
Her antenatal period was uneventful and fetal profile were normal throughout her pregnancy. She was planned for elective caesarean section. Pregnancy was terminated at 39 (EDD was on 07.07.2020) completed weeks of gestation by Cesarean section on 02.07.2020 and a live healthy

male baby was delivered. Intraoperatively a right sided ovarian lump was seen, right sided fallopian tube was absent. The lump was not adherent to adjacent structures and there was no free fluid in the peritoneal cavity. Right sided oophorectomy was done and specimen was sent for histopathological study. The left sided fallopian tube and ovary was normal.

Soft tissue specimen of right ovarian cyst measured 6x5cm^2. On cut section, the cyst contained yellowish solid material. Microscopic examination showed serous cystadenoma with ovarian stroma infiltration with foamy histiocytes, inflammatory cells and multinucleated giant cells. The tissue specimen was also subjected to special stains like Acid fast stain, PAS and GMNS stain and was negative for microorganisms thus ruling out Tuberculosis and fungal infection. Based on the unique pathological and histological features, xanthogranulomatous oophoritis was confirmed. The post-operative period was uneventful and on 6 months follow up patient was asymptomatic and her baby was doing well.

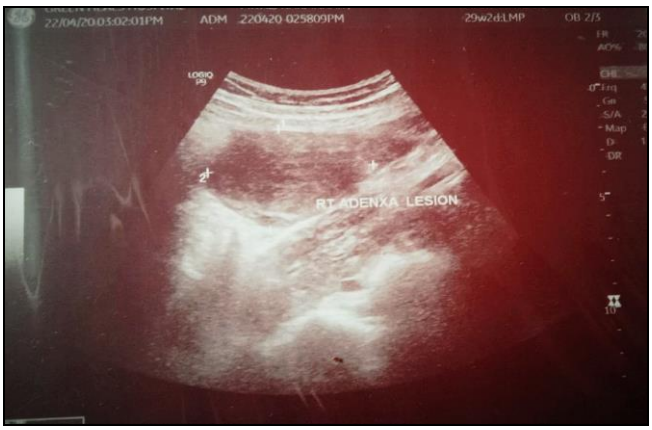

Fig 1: USG showing the right adnexal mass lesion

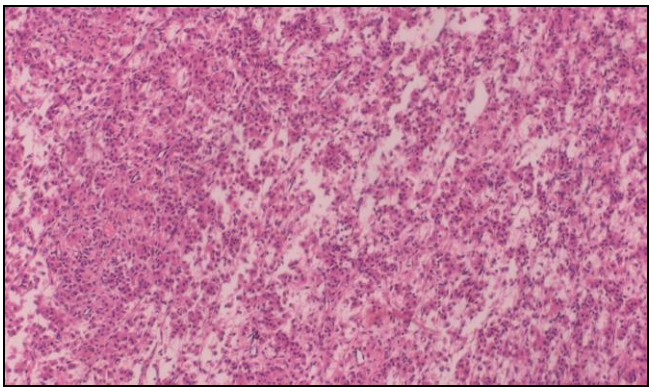

Fig 2: Microscopic low power view

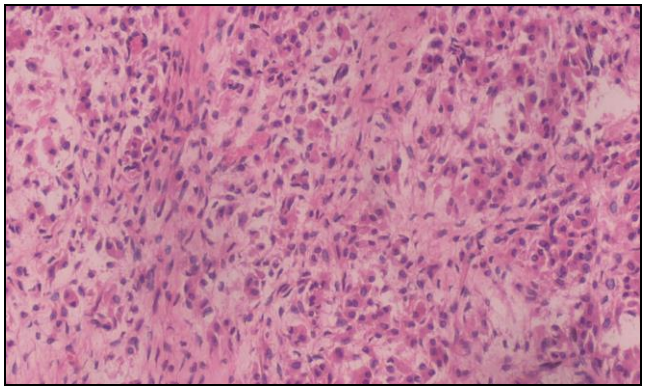

Fig 3: High Power 20x microscopic view

3. Discussion

Xanthogranulomatous inflammation of the female genital tract is very rare and if involved is confined to the endometrium [4]. It was first described by Kunakemakorn *et al*. in his first case of xanthogranulomatous inflammation of serosa of uterus, left fallopian tube, and ovary in his report of inflammatory pseudotumor in the pelvis in 1976 [5]. The youngest case was reported by Harshawardhan *et al*. in 2015 of a 2 year old female child with right sided xanthogranulomatous salpingo oophoritis [6].

The etiopathogenesis of xanthogranulomatous inflammation still remains unclear. Theories like abnormality in lipid metabolism, infection, ineffective antibiotic therapy and ineffective clearance of bacteria by phagocytes are proposed [7] Infection with organisms like Proteus, E coli and Bacteroides fragilis, Actinomyces and Staphylococcus aureus are reported as probable causative organisms in studies [8, 9] Uterine artery embolization, gloves dusting powder and altered lipid metabolism are also hypothesized to cause the pathology [10, 11, 12, 13]. Association with inadequately treated pelvic inflammatory disease Endometriosis, Intrauterine device use has also been reported [10].

Sometimes internal organ bleeding and obstruction may predispose to infection, tissue necrosis, followed by the release of cholesterol and other lipids and resultant in phagocytosis by macrophages and formation of foam cells [7] The previous history of tubal ectopic pregnancy followed by salpingectomy in our case might had been a predisposing factor for PID and tissue necrosis which might have led to such changes.

The condition very closely mimics Malakoplakia, where the foamy histiocytes have eosinophilic granular cytoplasm (Von Hansemann histiocytes) and show the cytoplasmic concentric calcific bodies known as Michaelis-Gutmann bodies which are absent in xanthogranulomatous inflammation [8] Immunohistochemical stains help in establishing the diagnosis, including CD 68 for histiocytes, CD 20 for B lymphocytes and CD 3 for T lymphocytes. Both the conditions may share a common pathogenesis [4].

Xanthogranulomatous oophoritis are often confused with ovarian malignancy because of similar clinical and radiological imaging findings. One case is reported in combination with ovarian serous cystadenoma by Anant M *et al*. in 2020 [7]. However no case has been reported with pregnancy. In our case, serous cystadenoma with xanthomatous oophoritis was present in pregnant state. Ovarian cysts are found in 4.1% of second-trimester and third-trimester obstetric sonographic examinations [14]. Of these, neoplasms constitute a significant number and most are benign. The types of ovarian cyst most frequently encountered in the pregnant women were dermoids and cystadenoma [15].

4. Conclusion

Serous cystadenoma is a bening neoplastic change of ovary and the most common benign ovarian neoplasm whereas xanthogranulomatous oophoritis is a chronic inflammatory change of the organ. Literature review shows no realtion in the etiopathogenesis of both. The pathogenesis of xanthogranulomatous change of ovary is not related to pregnancy as well. However, this case is unique because the patient had a history of same sided ectopic pregnancy as that of the ovarian mass (right side) for which she underwent right sided salpingectomy. This might have predisposed to infection and chronic inflammation which might have lead to xanthogranulomatous changes of ovary. We are reporting this

case because of this was a unique finding along with pregnancy and a neoplasm.

5. References

1. Bhatnagar K, Narang V, Garg B, Sood N. Xanthogranulomatous Oophoritis: A Rare Case Report. Iran J Pathol 2018;13(3):372-376. PMID: 30636962; PMCID: PMC6322522.
2. Yulia Gray, Peter Libbey N. Xanthogranulomatous Salpingitis and Oophoritis: A Case Report and Review of the Literature. Arch Pathol Lab Med 2001;125(2):260-263.
3. Jung SE, Lee JM, Lee KY, Han KT, Hahn ST. Xanthogranulomatous oophoritis: MR imaging findings with pathologic correlation, American Journal of Roentgenology. 2002; 178(3):749-751.
4. Patel KA, Kinalee CP, Bimal P, Lanjewar DN. Xanthogranulomatous salpingo-oophoritis: a case report and review of literature. International Journal of Reproduction, Contraception, Obstetrics and Gynecology 2020;9:43-16. 10.18203/2320-1770.ijrcog20204334.
5. Kunakemakorn P, Ontai G, Balin H. Pelvic inflammatory pseudotumor: a case report, American Journal of Obstetrics and Gynecology 1976;126(2):286-287.
6. Tanwar H, Joshi A, Wagaskar V, Kini S, Bachhav M. Xanthogranulomatous Salpingo-oophoritis: The Youngest Documented Case Report. Case Rep Obstet Gynecol 2015, 237-250. Doi: 10.1155/2015/237250. Epub 2015 May 31. PMID: 26114000; PMCID: PMC4465692
7. Anant M, Sinha K, Agarwal M, Sinha HH, Bhadani P. Serous cystadenoma of ovary with xanthogranulomatous oophoritis: combination of benign ovarian tumour with chronic inflammatory non-neoplastic pseudo-tumour-a rarity; International Journal of Reproduction, Contraception, Obstetrics and Gynecology Anant M *et al.* Int. J Reprod Contracept Obstet Gynecol. 2017;6(4):1682-1685.
8. Khan B, Aziz AB, Ahmed R. Case of Xanthogranulomatous Oophoritis. J Ayub Med Coll Abbottabad. 2017;29(1):162-164. PMID: 28712201.
9. Hota BM, Bakshi K, Movva N, Pandirla IS. Xanthomatous oophoritis, a rare pathology: case report with review of literature International Journal of Reproduction, Contraception, Obstetrics and Gynecology Hota BM *et al*. Int J Reprod Contracept Obstet Gynecol 2020;9(8):3486-3489.
10. Wlodarski FM, Trainer TD. Granulomatous oophoritis and salpingitis associated with Crohn's disease of the appendix. Am J Obstet Gynecol 1975;122:527-8.
11. McEntee GP, Goughlan M, Corrigan T, Dervan P. Pelvic inflammatory pseudotumor: problems in clinical and histologic diagnosis: case report. Br J Obstet Gynaecol 1985;92:1067-9.
12. Singh UR, Revathi G, Gita R. Xanthogranulomatous oophoritis: an unusual complication of typhoid. J Obstet Gynaecol 1995;21:433-6.
13. Singh N, Tripathi R, Mala YM, Arora S. Xanthomatous oophoritis following uterine artery embolisation: Successful conservative surgical management with favourable outcome. Case Reports 2013, bcr20130-10184.
14. Hill LM, Connors-Beatty D, Nowak A, Tush B. The role of ultrasonography in the detection and management of adnexal masses during the second and third trimesters of pregnancy. Am J Obstet Gynecol 1998;179(3):703-7.
15. Ross EK, Kebria M. Incidental ovarian cysts: When to reassure, when to reassess, when to refer Cleve Clin J Med 2013;80(8):503-14