Xanthogranulomatous Oophoritis – A Case Report

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ABSTRACT

Xanthogranulomatous inflammation is an uncommon form of chronic inflammation that is destructive of normal tissue in affected organs. Most commonly affected organs are kidney and gallbladder followed by anorectal area, bone, stomach and testis. This inflammation most commonly affects the endometrium in the female genital tract but involvement of vagina, cervix, fallopian tube and ovary may also occur. Only 16 cases have been reported in literature involving the ovary.

We report a case of 25 year old pregnant women presented with pain abdomen since 4 days at 35 weeks of gestation. Emergency LSCS was done along with left ovarian cystectomy. Histopathological examination revealed xanthogranulomatous oophoritis. This case is of interest in view of its rarity also due to its clinical and radiological resemblance to neoplastic lesion.

Keywords: Xanthogranulomatous oophoritis, oophorectomy, chronic oophoritis

INTRODUCTION

Xanthogranulomatous inflammation is an uncommon form of chronic inflammation in which the affected organ is destroyed and is replaced by large number of lipid-containing macrophages with an admixture of lymphocytes, plasma cells and multinucleated giant cells.[1] Most commonly affected organs are kidney followed by gall bladder.[1]

Other organs in which xanthogranulomatous inflammation has been reported are stomach, anorectal area, bone, urinary bladder, testis, epididymis and female genital tract.[4] Only 16 cases have been reported in the literature involving the ovary,[2,4] which will clinically presents as mass in the pelvic cavity and mimics an ovarian tumour clinically, radiologically and macroscopically.

CASE REPORT

A 25 year old G2P1L1 pregnant women presented with lower abdominal pain since 4 days at 35 weeks gestation. Physical examination showed tenderness in left iliac fossa. Ultrasound scan revealed 14x7x6 cm size biloculated hypoechoic mass suggestive of cyst[Figure 1]. Patient underwent emergency LSCS along with left ovarian cystectomy. Intraoperatively dense adhesions with omentum, bowel wall, uterus posterior wall and peritoneum were noted. Cyst wall rupture yielded 400ml of thick purulent fluid which was sent for culture and reported as positive for Escherichia coli.
We received a specimen of ovarian mass measuring 12x7x5cms. Gross examination showed grey brown to grey white nodular cystic mass [Figure 2]. Cut surface was partly solid and cystic with multiple loculae and yellowish solid areas [Figure 3]. Histologically sections from solid areas show dense infiltration of inflammatory cells [Figure 4], predominantly composed of foamy histiocytes [Figure 5] neutrophils, lymphocytes and occasional multinucleated giant cells and areas of necrosis. There was no evidence of malignancy in the sections studied.

**DISCUSSION**

Xanthogranulomatous inflammation is a form of chronic inflammation that is destructive to the normal tissue of affected organs.[3] Xanthogranulomatous inflammation is...
a well documented entity in gall bladder and kidney. However, Xanthogranulomatous inflammation of the female genital tract is unusual and essentially limited to the endometrium. Only a few cases involving the ovary, fallopian tubes and vagina have been reported.⁶ According to the literature 16 cases involving the ovary have been reported until 2012.⁶ Kunakemakorn was the first to describe xanthogranulomatous inflammation of serosa of the uterus, left fallopian tube and ovary in his report of inflammatory pseudotumor in the pelvis in 1976.⁷ The average age of occurrence is 31 years and the youngest case reported was of 18 years.

Xanthogranulomatous oophoritis (XGO) has an uncertain etiopathogenesis. Various factors implicated are chronic bacterial infection with E.coli, Proteus and Staphylococcus aureus, inadequate antibiotic therapy, endometriosis, intrauterine device and Inborn errors of lipid metabolism in macrophages.⁸ In the present case no predisposing factors were identified except the history of chronic pelvic inflammatory disease. Chronic or recurrent forms of ovarian involvement by pelvic inflammatory disease may take the form of chronic periophoritis with peri ovarian and tubo ovarian adhesions as seen in the case of our patient.

Chronic ovarian abscess may result in a solid tumor like mass, variably designated as ovarian xanthogranuloma, Xanthogranulomatous oophoritis, or inflammatory pseudotumor.⁹ XGO presents clinically as mass in the pelvic cavity and invades the surrounding tissues which can be misdiagnosed as neoplastic lesion and this may also be due to the rarity of the condition.¹⁰,¹¹ The involved ovary in XGO is replaced by a solid, yellow, lobulated mass characterised macroscopically by a well circumscribed, solid, yellowish, lobulated mass and can also present with cystic lesion at times due to liquefactive necrosis, microscopically by foamy histiocytes (xanthoma cells) admixed with multinucleated giant cells, plasma cells, fibroblasts, neutrophils, foci of necrosis and fibrosis.¹²,¹³ The yellowish coloration of this condition is due to the presence of foam cells.¹⁴,¹⁵

Chronic infection leads to tissue necrosis and continuously releases cholesterol and other lipids from dead cells, these cellular components are phagocytosed by macrophages, leading to xanthomatous process is also a possible explanation of this entity.¹⁶,¹⁷

Clinical presentations include anemia, anorexia, fever, menorrhagia and pain abdomen. Gynaecological examination reveal adnexal mass with tenderness and radiologically mimics ovarian tumor.¹⁸ In our case, patient has presented with recurrent pain abdomen complicating pregnancy. Shukla et al. have reported a case of XGO associated with primary infertility and endometriosis.¹⁹ Singh et al. have reported premature ovarian failure as a rare sequelae of XGO.¹⁰ Punia et al. have reported a case of XGO and salpingitis as a late sequelae of inadequately treated staphylococcal pelvic inflammatory disease.¹¹ Cases of xanthogranulomatous inflammation of ovary with ovarian hemangioma,¹² secondary to diverticulitis,¹³ association with endometriosis and uterine leiomyoma,¹⁴ secondary to talcum powder,¹⁵ presenting as an unusual complication of typhoid and following uterine artery embolization have been reported.

Treatment of choice for XGO is oophorectomy. Though cystectomy was attempted in our case during second trimester of pregnancy the recurrent ovarian mass was probably due to inadequate antibiotic therapy. Awareness of this inflammatory lesion is important to the pathologist and treating surgeon to prevent extensive surgery and also over diagnosis as malignancy.

CONCLUSION

Xanthogranulomatous oophoritis is a rare inflammatory lesion involving the ovary which usually presents with pelvic mass and abdominal pain. The clinical and radiological features may mimic an ovarian neoplasm, so it must be considered in the differential diagnosis of ovarian tumors. Since XGO is usually associated with pelvic inflammatory disease [PID], endometriosis, intrauterine death etc., these patients should be followed up closely. Although a correct diagnosis is chiefly made through histology, a suggestive preoperative diagnosis of XGO could lead to less radical surgery.

CONFLICT OF INTEREST:

The authors declared no conflict of interest.

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REFERENCES


