

BILATERAL XANTHOGRANULOMATOUS OOPHORITIS AND SALPINGITIS: A RARE CASE REPORTShashi Kant Kumar^{1*}, Ruchi Sinha², Kaushal Kumar³ and Shuchismita⁴^{1, 3, 4}Senior Resident, ²Associate Professor, Department of Pathology and Lab Medicine, All India Institute of Medical Sciences, Patna.***Corresponding Author: Shashi Kant Kumar**

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Article Received on 15/03/2017

Article Revised on 05/04/2017

Article Accepted on 26/04/2017

ABSTRACT

Xanthogranulomatous inflammation of ovary and fallopian tube is rare type of chronic inflammation and few are reported in the english literature. The affected organ is destroyed and replaced by lipid laden (foamy) histiocytes, admixed with lymphocytes, plasma cells, neutrophils, sometimes multinucleated giant cells and coagulative necrosis. Though it is very rare with preoperative clinical and radiological features mimic ovarian neoplasm, xanthogranulomatous oophoritis should be kept in mind while treating a patient with tubo-ovarian mass to avoid unnecessary radical procedure. We report a case of bilateral xanthogranulomatous oophoritis with salpingitis in a post menopausal women diagnosed clinically as ovarian neoplasm.

KEYWORDS: Bilateral, xanthogranulomatous, oophoritis.**INTRODUCTION**

Xanthogranulomatous inflammation is a form of chronic inflammation that usually leads to destruction of normal tissue of the affected organ.^[1] The most commonly affected organ is kidney followed by gall bladder.^[2] Other organs in which xanthogranulomatous inflammation has been reported are stomach, urinary bladder, endometrium, ovary and fallopian tube, testis, and epididymis.^[1, 3] Xanthogranulomatous inflammation of the female genital tract is very rare and mostly limited to the endometrium.^[4] Only a few cases involving the ovary and fallopian tube have been reported.^[1, 4] Clinical features, radiological appearance and gross features of xanthogranulomatous lesion of the ovary can mimic neoplastic lesion and lead to erroneous diagnosis.^[5] We report a case of bilateral xanthogranulomatous oophoritis with salpingitis in a 62 years old female.

CASE REPORT

A 62 year obese female presented to gynaecology outpatient department with chief complains of pain lower abdomen, white discharge per vaginum and swelling over midline scar on anterior abdominal wall with intermittent fever since several months. She had seven living children with history of sterilization about 20 year back. She was a known hypertensive on antihypertensive drug for 15 years. On physical examination, a pelvic mass measuring 8x8 cm was noticed with mild tenderness and midline incisional hernia in the anterior abdominal wall. Per speculum examination, revealed cervical erosion and a vague mass in the left adnexa. Ultrasonography of abdomen showed antverted atrophic

uterus, left ovarian cystic mass, normal right adnexa and midline incisional hernia. A contrast enhanced computer tomography scan of abdomen disclosed cystic mass in left adnexal region measuring 10x9x7 cm which was thought to represent either an ovarian cyst or cystic neoplasm adherent to omentum.

Routine haematological investigation showed mild anaemia with increased ESR (40 mm/ hr). Renal function test, liver function test, urine routine examination was within normal limits. Cervical smear examination was reported as negative for intraepithelial lesion or malignancy.

Tests for tumour markers like carcinoembryonic antigen (CEA), alfa fetoprotein (AFP), CA-125 were within normal limits. Based on the above findings a clinical diagnosis of ovarian neoplasm was made and the patient was posted for exploratory laparotomy. The patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy, segmental omental tissue resection and hernioplasty. The excised specimen was sent for histopathological examination.

PATHOLOGIC FINDINGS

The specimen received consisted of uterus with attached right adnexa, stump of cervix, left ovarian cyst, left fallopian tube and omental tissue. The uterus measured 5.5x5x2 cm, left ovarian cyst measured 7x2.5x1 cm and left fallopian tube measured 2.5x0.5 cm. The right ovary measured 2.5x2x1 cm and right fallopian tube measured 4x0.8 cm. The largest omental tissue measured 16x6x5

cm. The external surface of uterus, cervix, right adnexa, left fallopian tube and omentum were unremarkable. Outer surface of the left ovarian cyst showed large areas of pale yellow discoloration with foci of haemorrhage (figure -1), cut section revealed a unilocular cyst filled with thick white coloured fluid with no solid area identified. The cut section of right ovary was solid with pale yellow areas (figure-1). Serial sections of right fallopian tube show dilated lumen with inspissated yellow material inside. Histopathological examination of left ovarian cyst wall revealed destruction of ovarian stroma and replacement by inflammatory cells comprising sheets of foamy histiocytes, lymphocytes, plasma cells, few polymorphs and areas of necrosis (figure- 2). Sections from the right ovary and right fallopian tube also revealed similar microscopic features (figure-3 4). These findings were diagnostic of xanthogranulomatous inflammation of bilateral ovaries and right fallopian tube.

Figure legend



Figure 1: Gross of the uterus with bilateral adnexa. The left ovary is enlarged with areas of yellow discoloration. Cut section of the right ovary revealed solid pale yellow areas (arrow).

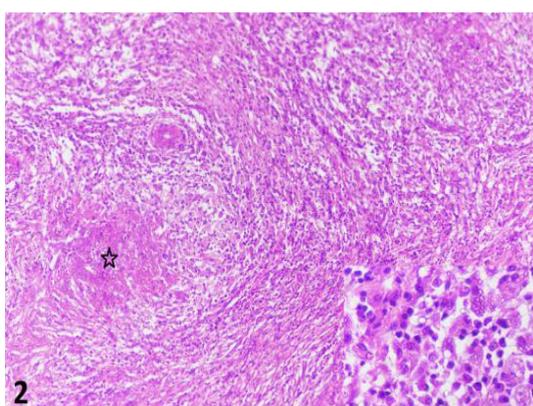


Figure 2: Section from left ovarian cyst wall shows destruction of ovarian stroma and dense infiltration of foamy histiocytes, lymphocytes, plasma cells and polymorphs with an area of necrosis (star). Inset show high power view.

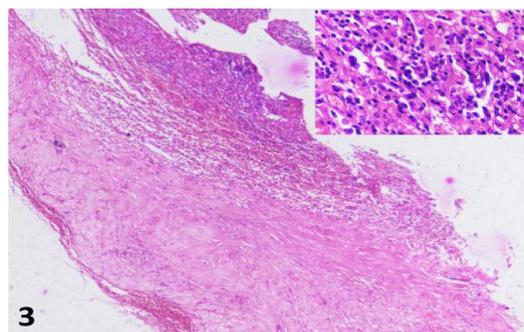


Figure 3: Section from right ovary shows destruction of ovarian tissue by dense chronic inflammation. Inset show high power view.

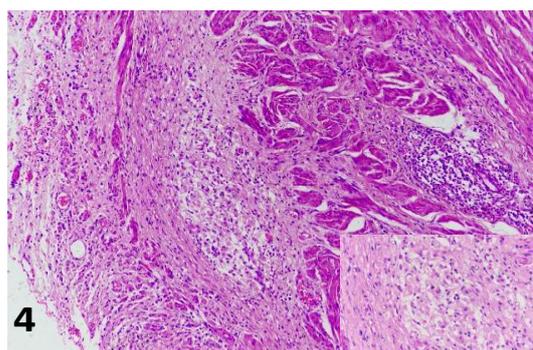


Figure 4: Section from right fallopian tube shows infiltration of foamy histiocytes, lymphocytes, plasma cells and polymorphs in between muscles layer. Inset show high power view.

DISCUSSION

Kunakemakorn was first to describe xanthogranulomatous inflammation of serosa of uterus, left fallopian tube and ovary in his report of inflammatory pseudotumour in the pelvis in 1976.^[6]

Only 16 cases of xanthogranulomatous inflammation involving the ovary and fallopian tube have been reported till date and with very few cases from Indian literature.^[7, 8] According to our search, all the reported cases are of unilateral involvement of ovary.

The etiopathogenesis of this disease is unclear but proposed factors include chronic bacterial infection, inefficient or inappropriate antibiotic therapy and ineffective clearance of bacteria by the phagocytes and abnormality of phagocytosis in macrophages.^[9] Gram negative or anaerobic bacterial genitourinary tract infections like *Bacteroides fragilis*, *Escherichia coli*, *Proteus vulgaris*, and *Salmonella typhi* are considered in the pathogenesis of xanthogranulomatous inflammation.^[1] Among these the most accepted theory is infection by *Escherichia coli* and other anaerobic bacteria which is supported by clinical evidence of infection and growth of bacteria from affected tissue by culture.^[10] Singh et al described xanthogranulomatous oophoritis as complications of typhoid.^[11]

Majority of the cases of xanthogranulomatous oophoritis belongs to age range 23-72 years (mean age 32 year) with the youngest case reported in 2 year female.^[12] The clinical presentations of xanthogranulomatous oophoritis include fever, abdominal pain, abdominal mass, menorrhagia, anaemia. Physical examination usually reveals pelvic mass with tenderness. Laboratory tests can show increased erythrocyte segmentation rate and white blood cell count.^[1, 9] Grossly, the affected ovary is enlarged, solid with yellowish appearance and occasionally cystic due to liquifactive necrosis. The inflammatory process may extend to the adjacent organ and pelvic peritoneum resulting in adhesion.^[7] Clinically and radiologically these features mimic malignancy.

Differential diagnosis of xanthogranulomatous lesion includes both inflammatory and neoplastic condition. Inflammatory conditions include malakoplakia and infections like tuberculosis and actinomycosis. Malakoplakia is an inflammatory disease which occurs chiefly in urinary system while xanthogranulomatous inflammation occurs mainly in the genital system.^[13] Malakoplakia shows intracytoplasmic concentric calcified bodies (Michaelis- Gutmann bodies) which are absent in xanthogranulomatous disease.^[14]

Infections can be ruled out by culture and special stain for the causative organism. Neoplastic condition should be ruled out by physical examination of the patient, radiology and blood investigation for tumour markers.

Treatment of choice for xanthogranulomatous oophoritis is oophorectomy.^[15] Although a correct diagnosis is made on the basis of histopathology, a suggested preoperative diagnosis of xanthogranulomatous oophoritis could lead to less radical surgery and post operative morbidity. Patient with inflammatory disease like endometriosis, pelvic inflammatory disease and intra uterine contraceptive device (IUCD) user kept on follow up due to their close association with xanthogranulomatous salpingo-oophoritis.

CONCLUSION

Bilateral xanthogranulomatous oophoritis and salpingitis is very rare in post menopausal women with clinical and radiological features closely mimic ovarian neoplasm, correct diagnosis and awareness of this entity is vital to prevent misdiagnosis as an adnexal neoplasm and avoid radical surgery.

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