
CASE REPORT**Xanthogranulomatous Oophoritis- A Rare Inflammatory Lesion***Mahesh H Karigoudar¹, Anirudha V Kushtagi^{1*}, Rashmi M Karigoudar², Anilkumar Sirasagi¹**¹Department of Pathology and ²Department of Microbiology, BLDE University Shri B M Patil Medical College, Hospital and Research centre, Bijapur-586103(Karnataka), India.*

Abstract:

Xanthogranulomatous inflammation is a special form of chronic inflammation that is destructive of affected organ. Xanthogranulomatous inflammation of ovary and fallopian tube is an uncommon entity and it is usually known to occur in the kidney and gall bladder. Patients with xanthogranulomatous oophoritis present with lower abdominal or suprapubic pain, fever, menorrhagia or vaginal bleeding, adnexal tenderness and a pelvic mass. This entity can be confused with ovarian tumour on clinical and radiological features. According to our search in the literature 16 cases of xanthogranulomatous oophoritis have been reported.

We report a 30 year old female, presented with a unilateral ovarian mass with clinical and radiological suspicion of ovarian tumour. Patient underwent panhysterectomy, grossly the right ovary was enlarged and on cut section it was solid with yellowish areas. Histopathological examination revealed massive infiltration of the ovarian tissue and fallopian tube by lipid-laden histiocytes admixed with mixed inflammatory cell infiltrate suggestive of xanthogranulomatous oophoritis. The case is of interest in the view of its rarity, also due to clinical and radiological resemblance to neoplastic lesion.

Key words: Xanthogranulomatous, inflammation, ovary, oophoritis, histiocytes

Introduction:

Xanthogranulomatous inflammation is a special form of chronic inflammation that is destructive to the tissue of affected organs. This is an uncommon process and usually known to occur in the kidney and gall bladder. Other organs in which xanthogranulomatous inflammation has been reported are as follows stomach, anorectal area, bone, urinary bladder, testis, epididymis, vagina, endometrium, ovary and fallopian tubes [1, 2]. Xanthogranulomatous inflammation of the female genital tract is unusual and limited to the endometrium. Xanthogranulomatous inflammation occurring in female genital tract affects endometrium, fallopian tubes or ovaries locally or entirely, which clinically presents with mass in the pelvic cavity and invades the surrounding tissues which can be misdiagnosed as neoplastic lesion and this may be also due to the rarity of the condition [1-4]. The clinical manifestations, radiological and macroscopic features of xanthogranulomatous oophoritis may mimic with ovarian tumour [2, 4].

Clinical features include lower abdominal or suprapubic pain, fever, menorrhagia, or vaginal bleeding, adnexal tenderness and a pelvic mass [1, 5]. According to our search in the literature 16 cases involving the ovary have been reported till 2012 [1, 4]. We report a case of xanthogranulomatous oophoritis in a 30 year old female because of its rarity.

Case report:

A 30 years old female presented with lower abdominal pain since long time. On examination pelvic mass was noted. Clinically diagnosis of pelvic inflammatory disease with right adenexal mass was made. Her cervical pap smears revealed non-specific cervicitis. Her haematological parameters were within normal limits except little rise in erythrocyte sedimentation rate (30mm/Hour). Sonographically right ovarian mass measuring 5x4x3cm was detected with suspicion of ovarian tumour. Patient underwent panhysterectomy.

Right ovary was enlarged and measuring 5x3.5x3 cm, cut section was solid with grey to yellowish areas. Other side of ovary was measuring 3x2x2cm and did not reveal any pathology on gross examination. Uterus and cervix were grossly unremarkable. The tubes on both sides were measuring 4cm each in length and right tube was slightly enlarged.

Microscopic examination of hematoxyline and eosin stained sections from right ovarian mass showed massive infiltration of the ovarian tissue by lipid-laden histiocytes known as Xanthoma cells admixed with mixed inflammatory cell infiltrate. Sections studied from tube of the same side revealed foci of similar microscopic features. Section with Zheel-Neelson stain was negative for acid fast bacilli, thus the diagnosis of xanthogranulomatous oophoritis was made.

Other side ovary, tube and endometrium were unremarkable but cervix revealed features of chronic nonspecific cervicitis.



Fig 1: Gross Enlarged Ovary with Grey to Yellowish Area on Cut Section

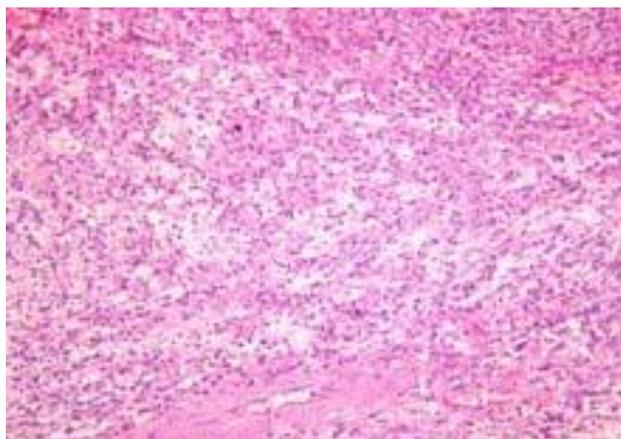


Fig 2: 100 X, Haematoxyline & Eosin, Mononuclear Inflammatory Cells, Foamy Macrophages and Ovarian Stroma

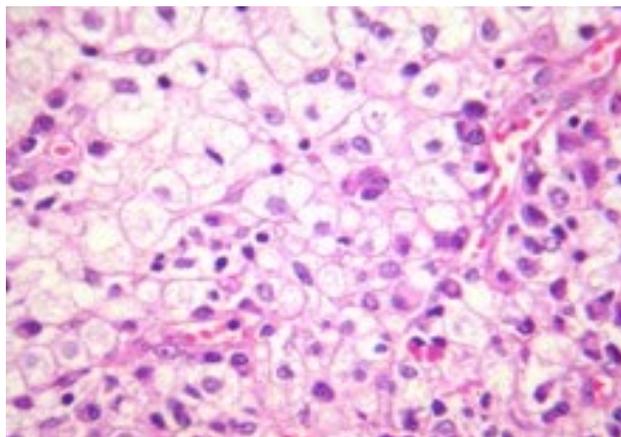


Fig 3: 400 X, Haematoxyline & Eosin, Mononuclear Inflammatory Cells & Foamy Macrophages.

Discussion:

Xanthogranulomatous inflammation of female genital tract is a rare and special type of chronic inflammation with destruction of the tissues of affected organs. Only 14 cases of xanthogranulomatous inflammation of ovary and fallopian tube have been reported in the literature till date [2,4,6]. The etiology and pathogenesis of this disease is unclear but many theories have been proposed, such as theory of infection, endometriosis, intrauterine contraceptive device, inborn error of lipid metabolism in macrophages and drugs (antibiotics), also combination of these factors may be responsible in its pathogenesis [1-7]. This lesion can also occur in patients with recurrent pelvic inflammatory disease as in our patient. Bacteria like *B. fragilis*, *E. coli*, *S. aureus* and *S. typhi* can be considered in the pathogenesis of xanthogranulomatous oophoritis [5]. In the present case no predisposing factors were identified except the history of chronic pelvic inflammatory disease.

Chronic infection leads to tissue necrosis and continuously releases cholesterol and other lipids from the dead cells, these cellular components are phagocytosed by macrophages, leading to xanthomatous process is also a possible explanation of this entity [8].

Clinical presentations include anemia, anorexia, fever, menorrhagia and pain abdomen. Gynecological examination can reveal adnexal mass with tenderness. Laboratory tests can show elevated erythrocyte sedimentation rate and increased white blood cell count [1,5]. Clinically and radiologically xanthogranulomatous inflammation may mimic ovarian tumor [1-6]. CT and MR imaging findings are usually nonspecific

for this condition. They are frequently misdiagnosed as malignant ovarian tumors due to CT and MR imaging appearances unusual for Tubo Ovarian Abscesses (TOAs) [3]. In our case, patient has presented with pain abdomen since more than six months and on examination lower abdominal mass has been detected. Sonography has revealed suspicion of ovarian tumor. Actinomycosis, tuberculosis and xanthogranulomatous inflammation of ovary are rare but are specific causes of TOAs, sometimes these can be misdiagnosed as ovarian neoplasms due to their unusual appearances on sonography, computed tomography and even on magnetic resonance imaging [3]. In our patient, on sonography ovarian tumour has been suspected. However, the correct diagnosis has required the help of histopathological study [2, 5]. In Xanthogranulomatous inflammation, the affected ovary can be replaced by a well circumscribed, solid, yellowish, lobulated mass and can also present with cystic lesion at times. In our case involved ovary has been found to be enlarged and solid with yellowish areas on cut surface. The yellowish color of this condition on gross examination is due to foam cells [1-6]. Microscopically it is characterized by a massive infiltration of the affected tissues by lipid-laden histiocytes known as Xanthoma cells admixed with mixed inflammatory cells consisting of lymphocytes, plasma cells and neutrophils [1, 2, 4, 5] and similar features have been seen in our case on microscopy. Because of the presence of foamy histiocytes, malakoplakia should be considered in the differential diagnosis of xanthogranulomatous inflammation [1, 2 and 4].

According to Wather, Malakoplakia and

xanthogranulomatous inflammation are identical chronic inflammatory diseases and which have the common pathogenetic pathway [2]. In xanthogranulomatous oophoritis, the cytoplasmic concentric calcified Michaelis-Gutmann bodies are not seen which can be seen in Malakoplakia [2, 4, 5]. In our case the Michaelis-Gutmann bodies have been not seen. Due to the rarity of the condition xanthogranulomatous oophoritis can often misdiagnosed as secondary lymphoma or leukemia also if the lesion is mainly with focal scattered lymphocytes. If the lymphocytes are scattering diffusely and foam cells are seldom, then there is a possibility of misdiagnosis of the condition as malignant small cell tumor with stromal luteinization. Sclerosing stromal tumor is also kept in the differential diagnosis when there are small amount of obvious fibrosis and foam cells. In our case, predominant infiltration with foamy histiocytes, few areas of sclerosis have been seen with mixed inflammatory cells.

Frozen section is also helpful in the diagnosis of xanthogranulomatous inflammation and further intra operative management [8]. Immunohistochemistry and the markers including CD68 (foam cells positive), CD3 (T lymphocyte marker), CD20 (B lymphocytes marker), κ and λ (both positive in polyclonal B lymphocytes) are also useful in establishing the diagnosis [2, 6].

In our case we have not studied immunohistochemistry, because the histopathological features confirmed the diagnosis. Surgery is the treatment of choice. Awareness of this inflammatory lesion can prevent extensive surgery and also over diagnosis as malignancy [5].

Conclusion:

Xanthogranulomatous inflammation of the female genital tract is an uncommon entity, clinical and radiological features of this entity may mimic ovarian tumour and hence it must be considered in the differential diagnosis of ovarian mass. Histopathological examination is essential for its diagnosis. Awareness of this entity is vital to avoid an incorrect and over diagnosis of malignancy. Correct diagnosis of this entity helps in the proper management of xanthogranulomatous inflammation.

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