CASE REPORT

Xanthogranulomatous Oophoritis, a Dilemma

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Received on: 02 January 2023; *Accepted on:* 30 January 2023; *Published on:* 31 July 2023

Abstract

Xanthogranulomatous oophoritis is a benign chronic inflammation causing massive destruction of involved organs. The disease is characterized by the infiltration of lipid-laden histiocytes or xanthoma cells along with giant cells, lymphocytes, and neutrophils. Diagnosis is often difficult as signs and symptoms are nonspecific and definitive evidence comes from histopathology.

We report a case of 27-year-old female presenting with infertility and an adnexal mass. Intraoperatively on laparoscopy, it was an ovarian abscess. Later on, through histopathology, it was diagnosed as xanthogranulomatous oophoritis.

Xanthogranulomatous inflammation in the ovaries is very rare, and when present, it can be a diagnostic challenge. The disease can mimic both benign and malignant ovarian masses, as such high degree of clinical suspicion combined with histopathology can aid in establishing the diagnosis.

Keywords: Oophoritis, Xanthogranulomatous, Xanthoma cells.

Journal of South Asian Federation of Obstetrics and Gynaecology (2023): 10.5005/jp-journals-10006-2245

Introduction

Xanthogranulomatous oophoritis is a rare benign chronic inflammation, causing massive destruction of the affected organ parenchyma by infiltration of lipid-laden histiocytes also known as xanthoma cells accompanied by giant cells, lymphocytes, and neutrophils. The disease is seen commonly affecting the kidneys, urinary bladder, stomach, gall bladder, testes, epididymis, and bones.[1] Involvement of the female genital tract is rare and mostly seen in the endometrium.[2] Ovarian involvement is very rare and when present, it may be misdiagnosed as an ovarian abscess or ovarian tumor due to its nonspecific signs and symptoms. The exact etiology is unknown, but many a time, it is seen associated with pelvic inflammatory disease, recurrent urinary tract infections (UTI), IUCDs, fibroids, endometriosis, inadequate antibiotic therapy, and an immunocompromised state.[1,2]

Case Description

A 27-year-old nulliparous female presented to Gynae OPD with dysmenorrhea and primary infertility for the last 4 years. Her menstrual cycles were regular with normal length and duration; however, it was associated with dysmenorrhea. She was being worked up for infertility in a local hospital, where on ultrasonoraphy (USG) examination, bilateral ovarian masses were diagnosed, and she was referred to a tertiary center. On examination, a vague mass was felt in the left vaginal fornix with tenderness; the uterus was normal in size with restricted mobility. Ultrasonography showed a 6.3 × 3.6 × 2.8 cm mass in the left adnexa and 1.5 × 1.4 cm mass in the right ovary, CA 125 was 35.6 IU/mL.

On provisional diagnosis of infertility with endometriosis, the patient was taken up for laparoscopy. A large mass of 6 × 8 cm was present in the left adnexa embedded in pouch of Douglas (POD) and densely adherent to bowel loops. Adhesiolysis was done, and it was separated from bowel loops; on rupture, it was filled with frank pus. A cystectomy was done and the cyst wall was sent for histopathological analysis. The right ovary was apparently normal with a follicular cyst (Fig. 1).

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How to cite this article: Ara A, Kumari K, Sharma C, *et al*. Xanthogranulomatous Oophoritis, a Dilemma. J South Asian Feder Obst Gynae 2023;15(3):357–358.

Source of support: Nil

Conflict of interest: None

Patient consent statement: The author(s) have obtained written informed consent from the patient for publication of the case report details and related images.

Histopathology examination revealed dense fibrosis and focal necrosis with the infiltration of ovarian stroma with foamy histiocytes, lymphocytes, and plasma cells suggestive of xanthogranulomatous oophoritis (Fig. 2). There was no evidence of endometriosis.

As our patient was nulliparous, we opted for conservative surgery. Later, when her histopathological findings came out for Xathogranulomatous oophoritis, she was under close surveillance with USG and Anti Mullerian hormone levels**.** In her follow-up period, the patient was doing fairly well and her AMH level was 1.8.

Discussion

Xanthogranulomatous oophoritis is a rare entity with very few cases reported in the literature. The presentation is commonly seen in females of reproductive age groups with an average being 31 years, but there are cases reported in extremes of ages also. Patients may present with pain, mild fever, anorexia, vaginal bleeding,

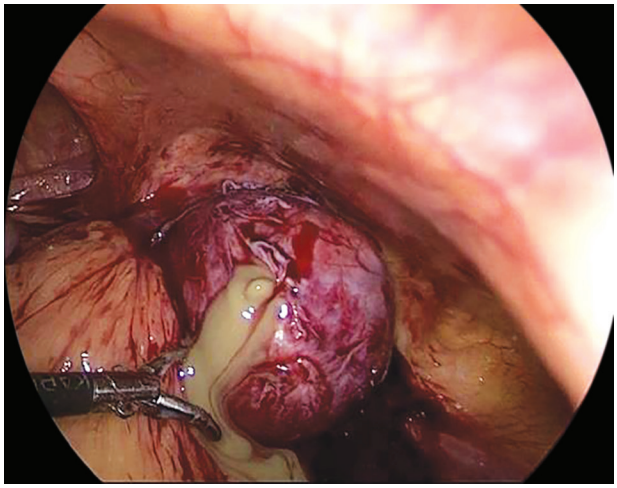

Fig. 1: Left ovarian mass filled with pus on laparoscopy

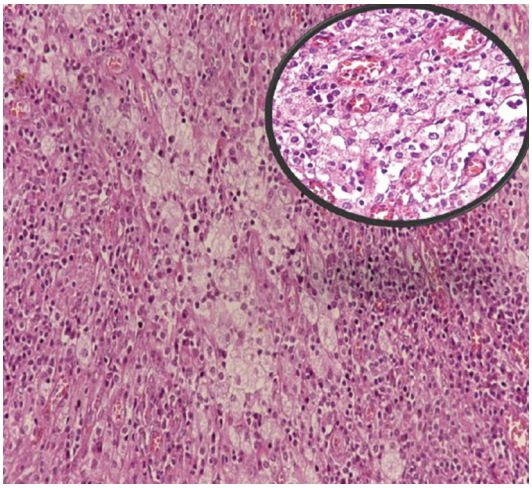

Fig. 2: Dense inflammation with foamy histiocytes (xanthoma cells magnified), sheets of plasma cells, and lymphocytes

dysmenorrhea, and adnexal masses. Signs and symptoms are nonspecific and often misleading. They are many a time mistaken as ovarian tumors, ovarian abscesses, tuberculosis, and sometimes endometriosis.

The exact etiology of xanthogranulomatous oophoritis is unknown, but multiple predisposing factors risk factors have been reported, such as long-standing pelvic inflammatory disease, recurrent UTI, intrauterine devices, endometriosis, lipid metabolism disorders, immunocompromised state, and inappropriate antibiotic therapy which sometimes lead to inadequate clearance of microbes resulting in persistent infection.[1,2] Commonly involved organisms are *Escherichia coli, Staphylococcus aureus*, group B streptococci, *Proteus vulgaris*, *Bacteriodes fragilis*, *Salmonella typhi*, Actinomyces, *Streptococcus* (Enterococcus) *faecalis*, *Streptococci viridans, Salmonella typhi*, and *Candida glabrata*.[2]

The exact diagnosis comes from histopathology, wherein the characteristic infiltration with lipid-laden histiocytes, foamy macrophages, admixed with multinucleate giant cells, lymphocytes, neutrophils, fibroblast, and plasma cells can be seen with areas of necrosis.[3]

In our case, the patient presented with infertility and adnexal mass, the preoperative diagnosis was endometriosis, the intraoperative diagnosis was an ovarian abscess, and postoperative histopathology came out as xanthogranulomatosis. There are very few case reports available in the literature and different etiologies and presentations have been described. In 2018, Bhatnagar et al. described a case of xanthogranulomatous oophoritis presenting as an ovarian abscess secondary to the sequel of chronic UTI with hydroureteronehprosis caused by Gram-positive cocci.[1] Elahi et al., in 2015, reported cases of xanthogranulomatous oophoritis presenting as ovarian neoplasm.[2]

Similar to our case, Shukla et al., in 2010, reported a case of xanthogranulomatous oophoritis mimicking endometriosis and presenting as infertility.[3]

In 2003, Punia et al. described a case of xanthogranulomatous salpingoophoritis after inadequately treated PID.[4]

As the disease is often mistaken and may mimic both benign and malignant ovarian neoplasm, analyzing the frozen section at the time of surgery can help in distinguishing benign from malignant pathology thus avoiding extensive surgery. Immunohistochemistry staining can be helpful to establish the diagnosis with a specific marker like CD68 in foamy histiocytes, CD20 in B cells, and CD3 in T cells. Special staining like Ziehl-Neelsen stain for acid fast bacilli (AFB) and PAS stain can be utilized to rule out tubercular and fungal infections.

Since the inflammatory process is intense, it tends to destroy the affected organ and dense adhesions are present with adjacent organs, as such the mainstay of treatment remains surgical excision which was also done in most of the cases.

Conclusion

Xanthogranulomatous oophoritis is a rare chronic inflammation that may sometimes be difficult to diagnose. The exact etiology remains uncertain, but some associated risk factors have been reported. The disease may mimic benign and malignant ovarian pathologies, such as abscesses and neoplasms on clinical and radiological findings. A high degree of clinical suspicion combined with histopathology aided with immunohistochemistry can help in ascertaining the diagnosis.

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