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Xanthogranulomatous oophoritis mimicking a dermoid cyst with ovarian torsion: A case report and review of the literature

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ABSTRACT

Xanthogranulomatous oophoritis (XO) is a rare pseudotumor representing a destructive chronic inflammatory process often mistaken for malignancy or tubo-ovarian abscess. Xanthogranulomatous inflammation is most commonly seen in the kidneys and gallbladder and very rarely affects the genitourinary system. Definitive treatment is with surgical removal of affected tissue.

This report presents the case of a 42-year-old woman with an 8 cm complex right adnexal cyst concerning for a dermoid cyst presenting with intermittent torsion. Final pathology after right salpingo-oophorectomy demonstrated xanthogranulomatous oophoritis.

This case is of clinical significance for distinguishing the condition from common benign pathology or cancer since the recommended surgical procedure is different than for a dermoid cyst or malignancy. Correct identification of the condition is crucial for appropriate treatment and to avoid unnecessary morbid procedures if the mass is mistaken for malignancy or future repeat surgery if mistaken for a dermoid cyst or other common benign condition. This case documents the presentation of xanthogranulomatous oophoritis masquerading as a dermoid cyst for a condition with very few reported cases worldwide.

1. Introduction

Xanthogranulomatous inflammation is a destructive inflammation characterized by the invasion of foamy macrophages, neutrophils, multinucleated giant cells, lymphocytes, and plasma cells that result in parenchymal obliteration. This specific type of chronic inflammation is most commonly observed in the setting of pyelonephritis or cholecystitis. Less commonly, it affects the stomach, testis, anorectal area, bone, urinary bladder, and epididymis [1]. The condition very rarely affects the female genitourinary system, but has been known to infiltrate the endometrium, resulting in xanthogranulomatous endometritis, causing destruction of the endometrium, pain, and abdominal distension [2]. However, fewer than 50 cases have been reported of xanthogranulomatous inflammation affecting the ovary or fallopian tube, with the vast majority reported in India. Pathogenesis and risk factors for this condition are poorly understood. Patients typically present with abdominal and pelvic pain, fever, vaginal bleeding, tenderness, and a pelvic mass [1]. These pseudotumors are commonly mistaken for malignancy, tubo-ovarian abscesses, or tuberculosis [3,4].

The present report describes a case of xanthogranulomatous oophoritis (XO) mimicking a dermoid cyst in a 43-year-old woman with clinical suspicion for ovarian torsion. Histopathology after laparoscopic salpingo-oophorectomy ultimately provided the diagnosis.

2. Case Presentation

A 42-year-old woman presented to clinic with stabbing right-sided pelvic pain that intermittently improved with rest and heat. Her past medical history was significant for celiac disease and microscopic colitis and surgical history included an appendectomy and cholecystectomy. She smoked daily and had a Mirena levonorgestrel intrauterine device (IUD) for contraception. A pelvic exam performed in clinic noted pelvic tenderness but no adnexal masses, so outpatient imaging was recommended to evaluate the position of her IUD.

A pelvic ultrasound demonstrated a 7.4 × 6.0 × 7.8 cm complex heterogeneously hypoechoic right adnexal mass with small echogenic foci (Fig. 1). No internal blood flow was observed on color doppler and the left ovary appeared normal. The lesion was given an O-RADS score of

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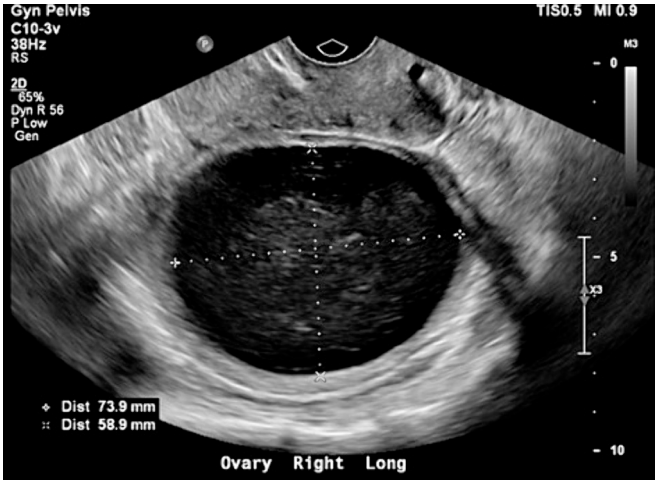

Fig. 1. Transvaginal sonogram of right adnexal mass.

3, which indicates a relatively low risk of malignancy (between 1% and 10%).

Upon follow-up, her pelvic exam demonstrated a large tense fluctuant mass occupying the posterior cul de sac, exquisitely tender with palpation of the right adnexa and right-sided rebound tenderness of the abdomen. No left adnexal tenderness was elicited. The large size of the complex O-RADS 3 mass with internal echogenic foci lacking blood flow on imaging increased suspicion for a dermoid cyst. She was admitted directly to the inpatient gynecology service for expedited workup of a suspected dermoid cyst with possible intermittent torsion.

A laboratory workup was notable for a CA-125 of 19 U/mL, a negative urine pregnancy test, and a normal CBC. Due to ongoing significant pain, the decision was made to proceed directly to the operating room for a laparoscopic right ovarian cystectomy and possible oophorectomy.

Intraoperatively, a normal uterus, bilateral fallopian tubes, and left ovary were observed. An approximately 8 cm cystic right ovarian mass with a smooth external surface was seen (Fig. 2). The mass was well vascularized, without evidence of torsion, and with minimal identifiable normal ovarian tissue. The surgeons proceeded with a right salpingo-oophorectomy and opportunistic left salpingectomy as the patient made clear she had completed childbearing. During manipulation of the ovarian mass, it was inadvertently ruptured, with intraabdominal spillage of thick yellow fluid. The abdomen and pelvis were copiously irrigated prior to closure.

Her postoperative course was uncomplicated and she was discharged home on postoperative day 1. Final pathology demonstrated the right ovary with prominent chronic inflammation consisting predominantly of histiocytes, lymphocytes, and plasma cells (Fig. 3). Although the ovary had a cystic appearance, true epithelial lining cells were not identified on H&E stains or highlighted on cytokeratin or CK8/18 stains. There was no evidence of malignancy. The inflammatory cells showed a mixture of CD68 stain positive histiocytes (Fig. 4) and CD3/CD20 positive lymphocytes. Bilateral fallopian tubes were without pathologic change.

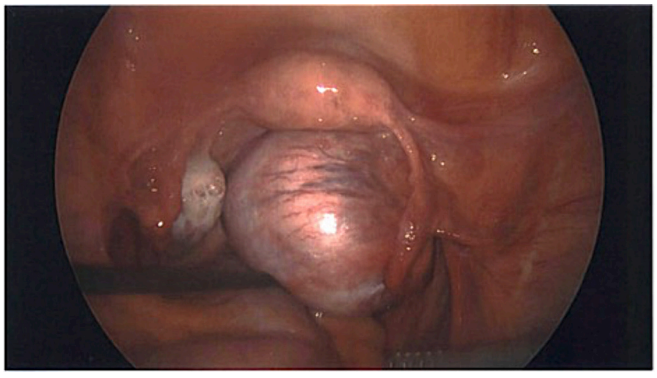

Fig. 2. Right fallopian tube and ovary. Intraoperative laparoscopic imaging of normal-appearing uterus, left fallopian tube and ovary, and abnormal mass encompassing right ovary.

3. Discussion

Adnexal masses in premenopausal women are common, with studies reporting prevalence as high as 34.9% [5]. Very few are diagnosed as malignant and the vast majority of malignancies of tubo-ovarian origin arise in postmenopausal women. Transvaginal ultrasound is the mainstay of diagnostic imaging for the evaluation of adnexal masses and is crucial for determining risk of malignancy for appropriate referral to gynecologic oncologists; however, it is not as reliable in women of reproductive age due to the lower prevalence of malignancy and higher prevalence of alternative diagnoses [6]. Xanthogranulomatous inflammation is a form of chronic inflammation most frequently documented in the kidneys and gallbladder and very rarely found in the female genital tract [1]. When xanthogranulomatous inflammation is found in the genital tract, the endometrium is the most common location, whereas it is much less common in the fallopian tube and ovary. There are fewer than 50 searchable documented cases of XO worldwide, with the majority presenting in India and Singapore. The condition is not well documented in the United States.

Of the published cases, the most common presentation of XO is abdominal pain, followed by fever, pelvic mass, abnormal uterine bleeding, and abnormal vaginal discharge [7]. The adnexal masses were most commonly mistaken for malignancy or tubo-ovarian abscess. XO

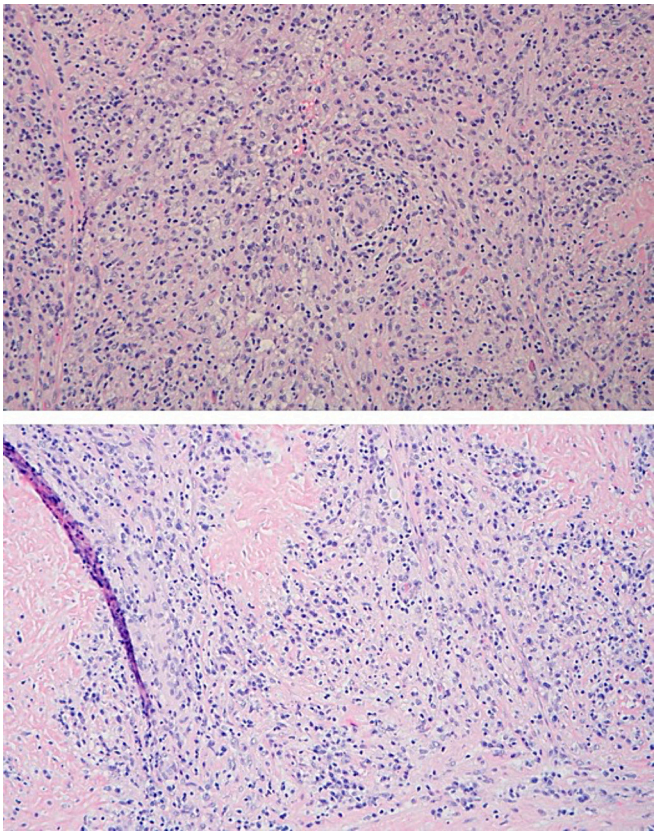

Fig. 3. H&E stains of right ovarian tissue with xanthogranulomatous oophoritis at 20× magnification.

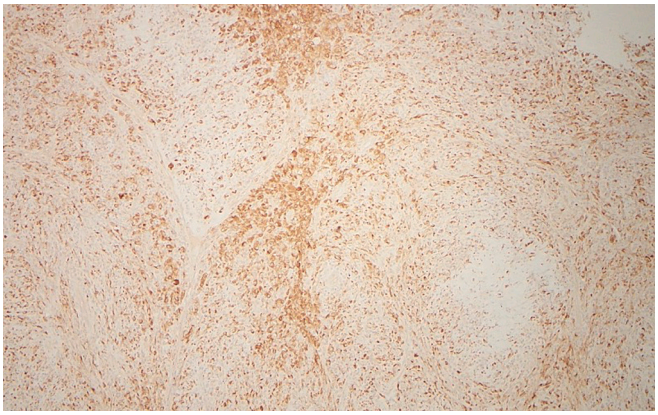

Fig. 4. Immunohistochemistry CD68 staining of right ovarian tissue demonstrating prominence of histiocytes characteristic of XO.

has also been reported with endometriosis, infertility, and bowel conditions [4,8]. Classic imaging demonstrates a large (5–15 cm) complex loculated mass, often with enhancing septa and solid and cystic components [8]. The pathology may involve both ovaries and/or the fallopian tubes [3]. Due to the rarity of XO, risk factors have not been well established in the literature; however, several factors associated with inflammation have been posited as risk factors, including chronic pelvic inflammatory disease (PID), current IUD use, and tobacco use [4]. This patient exhibited two out of three of these risk factors. Other etiologies postulated in the literature include insufficient antibiotic treatment of pelvic infection, endometriosis, inborn errors of lipid metabolism, or drug induced by antibiotics or lipid-containing drugs. Microorganisms suspected to play a part in the pathogenesis include *Bacteroides fragilis*, *Escherichia coli*, *Staphylococcus aureas*, and *Salmonella typhi* [3,8]. Pathologic examination after surgical excision is the only method of definitive diagnosis, but is challenging for pathologists. Use of immunohistochemical stains can assist in establishing the diagnosis, particularly markers for foam cells (CD68), T lymphocytes (CD3), B lymphocytes (CD20), and polyclonal B lymphocytes (κ and λ) [3].

Surgical management of adnexal masses in patients of reproductive age typically involves laparoscopic excision of the mass with ovarian preservation for benign indications, and staging procedures involving bilateral salpingo-oophorectomy, hysterectomy, and biopsies in cases of high suspicion for malignancy. XO is frequently misdiagnosed preoperatively as malignancy due to low familiarity with the condition and imaging which typically demonstrates a large solid-cystic complex adnexal mass sharing features with malignancy [3]. CA-125, a serological marker used to help identify epithelial ovarian cancer, can be elevated in the setting of XO [9], further confusing the two diagnoses. Frozen pathology is widely utilized in gynecologic oncology for intraoperative diagnosis and aids in the decision to proceed with the appropriate surgery for patients with gynecological cancer who do not have a preoperative histopathological diagnosis. A previous case series stresses the importance of preoperative biopsy or intraoperative frozen pathology in cases of XO vs malignancy as well to avoid unnecessary surgery, since several patients in reported cases received extensive staging surgeries for suspected malignancy that turned out to be XO, and were therefore unindicated [3]. For this reason, prior case reports encourage caution in proceeding with a staging surgery under suspicion of malignancy when xanthogranulomatous oophoritis could be the diagnosis [9].

In this case, however, the preoperative diagnosis could have resulted in inadequate surgical management. The recommended procedure for a premenopausal woman with a dermoid cyst is a cystectomy with preservation of ovarian tissue, whereas complete oophorectomy is recommended for xanthogranulomatous oophoritis due to the obliterative nature of the disease process. A report in the literature describes a suspected dermoid cyst that was determined to be both mature cystic teratoma and xanthogranulomatous oophoritis and the patient was suboptimally treated with a cystectomy [10]. A high degree of suspicion is essential for this rare disorder when there is low suspicion for malignancy in a premenopausal woman and risk factors for XO. In suspected cases, frozen pathology of a cystectomy specimen may be useful in making an intraoperative decision to proceed with cystectomy vs oophorectomy when ovarian preservation could risk the need for additional interval surgery.

4. Conclusion

Xanthogranulomatous oophoritis is a rare chronic inflammatory pathology diagnosed on histologic examination after surgical resection. Its etiology is unknown but thought to be secondary to chronic infection and inflammation. While the condition can be easily mistaken for malignancy on imaging, this case further demonstrates its capacity to mimic benign pathology. However, due to its frequent misdiagnosis as malignancy or pelvic inflammatory disease and the challenging nature of pathologic diagnosis, the prevalence of this disease process is likely underestimated [7,9]. Vigilance is key in patients who may have XO instead of a malignancy or a benign cyst in order to avoid surgical over- or under-treatment. In patients presenting with risk factors associated with XO, intraoperative frozen pathology may assist in the diagnosis and decisions regarding surgical planning.

Contributors

Carrie A Sibbald contributed to patient care, conception of the case report, acquiring and interpreting the data, drafting the manuscript, undertaking the literature review and revising the article critically for important intellectual content.

Laura G Cooney contributed to revising the article critically for important intellectual content.

Ross J Molot evaluated the pathologic specimen and made the histopathological diagnosis.

Daniel L Pellicer contributed to patient care, conception of the case report, and revising the article critically for important intellectual content.

All authors reviewed and approved the final submitted manuscript.

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Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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