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Xanthogranulomatous salpingo-oophoritis infiltrating into rectum

Pooja Agarwal, Ruquaya Mir, Pankaj Baweja

ABSTRACT

Xanthogranulomatous salpingo-oophoritis of the female genital tract is an extremely rare entity with less than 30 cases being previously reported in the literature. It is characterized by destruction of the normal tissue followed by replacement with inflammatory cells and foamy macrophages. The clinical characteristics closely resemble those of typical noncancerous and cancerous adnexal conditions, making diagnosis challenging. In this case, we describe a patient with a pelvic mass, elevated tumor marker levels, and a history of using an intrauterine contraceptive device. She underwent surgery with the suspicion of an adnexal tumor, but the pathological examination revealed it to be xanthogranulomatous salpingo-oophoritis.

Keywords: Adnexal mass, CA-125, IUCD, Malignancy, Xanthogranulomatous salpino-oophoritis

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INTRODUCTION

Xanthogranulomatous inflammation is an uncommon cause of chronic inflammation affecting various organs, leading to the destruction of the normal tissue in the affected organ. It is characterized by the accumulation of foamy macrophages intermixed with neutrophils, lymphocytes, and plasma cells along with multinucleated giant cells [1]. Xanthogranulomatous inflammation primarily targets the kidney, but it can also impact other organs like the gallbladder, stomach, anorectal region, bone, urinary bladder, testis, and epididymis. Xanthogranulomatous inflammation of the female genital tract is rare and if found is mostly limited to the endometrium [2]. Only a few cases involving the ovary and fallopian tube have been reported [2, 3]. The clinical symptoms, radiological appearance, and physical characteristics of xanthogranulomatous lesions in the ovary can resemble those of cancerous growths, potentially leading to misdiagnosis. After conducting an extensive review of the existing literature, we discovered that only 15 cases related to xanthogranulomatous oophoritis involving the fallopian tube or ovary have been documented, with an average patient age of 31 years [1]. Typically, these cases manifest as either a pelvic inflammatory condition unresponsive to antibiotics or as a mass that mimics pelvic cancer [3]. In this instance, we present a case of xanthogranulomatous salpingo-oophoritis in a patient with a history of intrauterine contraceptive device and elevated tumor markers, which initially appeared as if it were an ovarian malignancy. This condition can cause frozen pelvis and resection of various pelvic organs unless diagnosis is made preoperatively.

CASE REPORT

A 39-year woman, P2L2, was admitted with chief complaints of lower abdominal pain along with increased frequency of defecation since four months. Pain was insidious in onset, dull aching and mainly on the left side of abdomen, not associated with menstrual cycle and relieved on taking pain killers. Menstrual cycle was regular with normal flow. There were no other complaints. She was a known case of hypothyroidism, on medication, and no history of any addiction. She had history of two cesarean sections. Her history revealed that she was having in situ intrauterine contraceptive device (IUCD).

She was hemodynamically stable. The abdomen was soft, scaphoid, and non-tender, with previous cesarean Pfannenstiel scar and there was no palpable mass. The cervix visualized and was pulled up. On per vaginal and rectal examination, the uterus was normal. A firm, non-tender mass was palpated in the left side of the pouch of Douglas. The rectal mucosa was free of abnormalities.

Ultrasonography (USG) abdomen revealed heterogeneous hypoechoic solid cystic lesion measuring 8.1 × 6.8 × 5.3 cm in left adnexal region abutting posterior myometrium. Endometrial cavity has intrauterine contraceptive device (IUCD). Magnetic resonance imaging (MRI) pelvis revealed large solid lesion in midline measuring 12.2 × 11.5 × 11 cm contiguous to adjacent bowel loops.

Her complete blood count was within a normal range. Serum ovarian cancer marker (CA-125) was 74.60 U/mL (high). A staging laparotomy was performed, which revealed complex left tubo-ovarian mass measuring 7 × 6 × 5 cm and was densely adhered to the rectum and sigmoid colon with pelvic abscess. Anteriorly it was adhered to urinary bladder. The right ovary and fallopian tube seemed to be normal. The patient underwent a total abdominal hysterectomy with bilateral salpingo-oophorectomy with anterior resection and diversion ileostomy. She had an uneventful postoperative recovery and was discharged on the fifth postoperative day. Histopathological examination revealed that the pelvic peritoneum showed features consistent with abscess formation. Acute on chronic cervicitis with squamous metaplasia. Histopathological examination revealed proliferative endometrium with areas of abscess formation. Bilateral ovaries showed features of abscess formation with focal aggregates of foamy macrophages suggestive of bilateral salpingo-oophoritis. Periodic acid Schiff stain, gram stain, and acid-fast stains were negative for microorganism. Adhered anterior resection specimen showed extensive serosal exudates with areas of abscess formation and aggregates of foamy macrophages. Immunohistochemistry (IHC) foamy macrophages showed diffuse cytoplasmic positivity for CD68, which confirms the diagnosis of xanthogranulomatous salpingo-oophoritis (Figure 1).

DISCUSSION

Xanthogranulomatous inflammation of uterus, fallopian tube, and ovary was first described by Kunakemakorn in 1976 [3]. Till date, only 15 cases of xanthogranulomatous inflammation of the female genital tract are reported in literature out of which there are 7 cases with unilateral ovary, 5 cases with unilateral fallopian tube, 1 case with bilateral fallopian tube involvement, and only 2 cases with ovary and fallopian tube showing simultaneous involvement [1]. Thereafter, 13 cases of xanthogranulomatous salpingitis from the years 2003 to 2018 were reported by Chiesa-Vottero, in which most of the clinical presentations mimicked female genital tract malignacy [4]. Xanthogranulomatous salpingitis has been observed in individuals ranging from 21 to 75 years old, with an average age of 45 years [4, 5]. The condition has even been reported in a two-year-old patient (Tanwar et al., 2015) [6]. While the precise cause of xanthogranulomatous salpingitis remains unclear, it is linked to factors that lead to the formation of foam cells. Factors such as the presence of endometriosis and pelvic inflammatory disease, the use of intrauterine contraceptive devices, ineffective antibiotic treatment, or abnormal lipid metabolism are considered primary contributors to this condition [7, 8]. Infections by various microorganisms like *Escherichia coli*, *Proteus vulgaris*, *Bacteroides fragilis*, and *Salmonella typhi* have also been associated with this type of inflammation. It is thought that the colonization of the IUCD in the endometrial cavity is followed by the shedding of bacteria through the lumen of the fallopian tube [9]. Alternatively ovarian surface is exposed to bacteria at the time of ovulation, the corpus luteum becomes infected, producing an ovarian abscess.

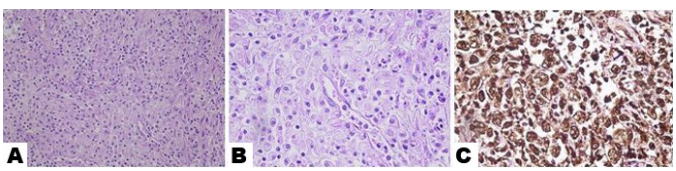

Figure 1: (A and B) Histology shows diffuse proliferation of foamy macrophages and inflammatory cells. (C) shows CD68 cytoplasmic positivity in foamy macrophages (IHC).

Microscopically, it is characterized by a massive infiltration by foamy histiocytes known as Xanthoma cells and mixed with inflammatory infiltrates of the affected tissues [10, 11]. Immunohistochemical stains CD68, CD3, and CD20 are helpful in establishing the diagnosis if any doubt is there in histomorphological examination.

The clinical scenario may vary but usually patients present with pain in the lower abdomen, fever, dysmenorrhea, dyspareunia, chronic pelvic pain, bleeding, poor appetite, mass in the abdomen, and/or infertility [3, 12].

In addition to non-neoplastic and neoplastic diseases, xanthogranulomatous salpingitis and oophoritis have a wide differential diagnosis. Infections including tuberculosis, fungal infections, and malakoplakia are the most common non-neoplastic illnesses, and they must be ruled out through cultural studies and special stains like the periodic acid Schiff stain, gram stain, and acid-fast stains. Due to its destructive nature and tendency to form masses, distinguishing this disease from malignancy is extremely challenging, if not impossible, both in terms of clinical symptoms and radiological findings. In our research, the patient displayed an adnexal mass and an elevated CA-125 level, which resembled a pelvic tumor. Consequently, surgery was performed with the suspicion of malignancy. However, it was a pathological surprise clinching with the diagnosis of bilateral xanthogranulomatous salpingo-

oophoritis. As a result, xanthogranulomatous salpingo-oophoritis is exceedingly difficult to detect preoperatively and is typically discovered after histological analysis due to its rarity and clinical characteristics that resemble pelvic organ tumors. In order to correctly diagnose the cause of inflammatory disorders and treat those that affect the female genital system, thorough consideration is therefore required [3].

CONCLUSION

Xanthogranulomatous salpingo-oophoritis is a rare condition that can exhibit clinical symptoms resembling those of common benign and malignant adnexal diseases. Therefore, it necessitates a heightened level of suspicion and awareness among clinicians, radiologists, and pathologists to facilitate early diagnosis and proper management, particularly in patients with pelvic inflammatory disease (PID), endometriosis, and intrauterine contraceptive device (IUCD) usage, among other factors.

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Author Contributions

Pooja Agarwal – Conception of the work, Design of the work, Acquisition of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ruquaya Mir – Design of the work, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Pankaj Baweja – Acquisition of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

Source of Support

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Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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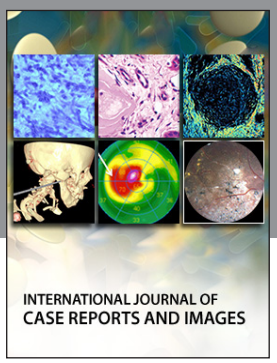

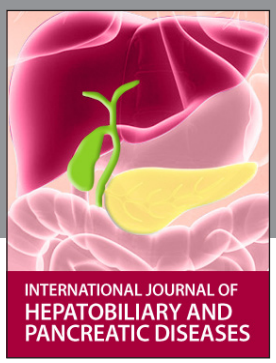

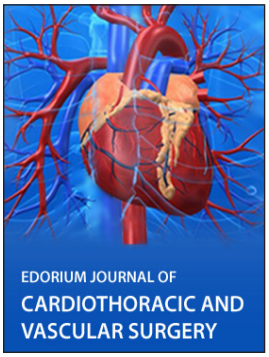

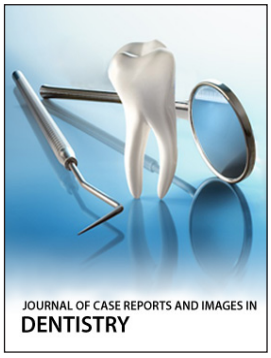

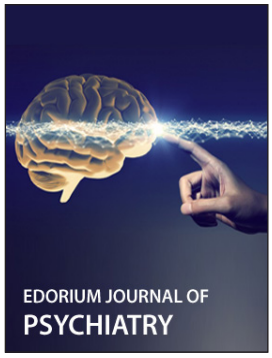

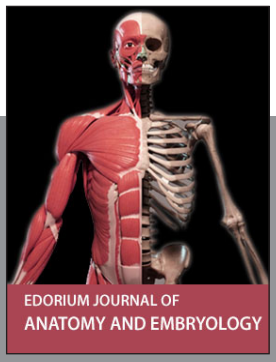

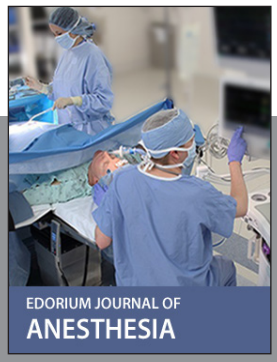

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