CASE REPORT

Xanthogranulomatous oophoritis: a diagnostic dilemma

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Introduction

Xanthogranulomatous oophoritis is a rare diagnosis in patients with ovarian mass. Patients with xanthogranulomatous oophoritis present with mass per abdomen with imaging suggestive of malignancy sometimes even invading surrounding structures. Tumour markers for ovarian malignancy serum Ca 125 values may be marginally raised. These common features of xanthogranulomatous oophoritis with malignancies of ovaries have given it a special name pseudotumour of ovary.

Case Reports

Case 1

Sixty-seven-year-old P3L3 lady menopausal for 10 years presented with history of pain abdomen on and off of 6-month duration relieved with analgesics, swelling in

lower abdomen of 3-month duration. Patient had retention of urine 2 months before the present consultation. She was catheterised and treated with antibiotics and analgesics, with clinical diagnosis of urinary tract infection. Since patient persisted to have retention of urine after removal of catheter, she was evaluated with imaging. Ultrasonography suggested a pelvic mass suggestive of malignancy. Patient was referred to our centre for further management.

Her general physical examination was normal. Per abdomen examination showed suprapubic mass with restricted mobility, measuring $15 \times 15 \times 10$ cm, no ascites and no hepatosplenomegaly. Investigations revealed haemoglobin 9.8 g%; Ca 125 –14.83 U/ml; CEA –0.78 U/ml; otherwise normal.

The sonologist described the mass as large rounded, cystic lesion measuring $11.6 \times 11.5 \times 12.4$ cm, sized arising from pelvis, with densely packed internal echoes, few membranes noted with in and foci of calcification. Ovaries were not separately seen. CT scan showed a lower abdomen and pelvic mass measuring $13.8 \times 10 \times 12.3$ cm, with thick and predominantly regular wall except for anterior and right lateral aspect which is thickened with enhancing soft tissue along this wall. There is significant extralesional fat stranding seen along this wall with tubular fluid collection extending laterally for a length of 8 cm. A thin focal streak of calcification was seen along the anterior wall. Long segment thickening of distal ileum and caecum adjoined the lesion for a length of 7.0 cm. Both ovaries were not seen. No significant lymphadenopathy was observed.

Patient was taken up for laparotomy. Intraoperative findings reveal uterus and both tubes normal and left ovary normal. Right ovary showed a thick-walled cyst containing about 1.5-L thick greenish pus with adhesion to bowel loops, uterus and omentum. Right ovary appendix was grossly normal. Patient underwent hysterectomy with bilateral salpingo oophorectomy with excision of the cyst with its contents. The part of omentum adherent to cyst was excised.

Histopathological examination confirmed xanthogranulomatous inflammation of the cyst arising from right ovary and also in the part of omentum adherent to it (Fig. 1).

Patient recovered well during postoperative treatment, and she is on follow-up for last 1 year.

Case 2

Forty-three-year-old P1L1 lady with regular menstrual cycles presented with C/o pain abdomen on and off with watery discharge per vaginum and loss of appetite for 15 days. Her general physical examination was normal except for pallor. Per abdomen examination showed no palpable mass, organomegaly or free fluid felt.

Fig. 1 Histological image of xanthogranulomatous inflammation with foamy macrophages and histiocytes

Fig. 2 CT scan image of case 2 showing thick-walled pelvic cyst with adherent bowel wall

Rectovaginal examination revealed a 10×8 cm mobile mass felt in right adnexa. No forniceal tenderness was observed. Parametrium and rectal mucosa were free. Investigation revealed haemoglobin 9.2 g%; tumour markers Ca 125–66.64 U/ml; CEA –3.23 ng/ml; otherwise normal. Ultrasonography showed uterus measuring $11.9 \times 6.9 \times 6.6$ cm with subserous and intramural fibroids. Left ovary showed complex cystic mass measuring 11.8×8.9 cm with septae and internal echoes (Fig. 2). Patient underwent laparotomy. Intraoperatively, uterus measured $12 \times 6 \times 5$ cm with posterior wall fibroid measuring 5×6 cm, and left-sided tubo ovarian mass measured $10 \times 8 \times 6$ cm buried in the broad ligament found. Hysterectomy with bilateral salpingo oophorectomy was done.

Histopathological examination confirmed left ovarian xanthogranulomatous oophoritis.

Discussion

Xanthogranulomatous oophoritis is a rare form of chronic ovarian inflammation. Inflammation with xanthogranulomatous changes contains lipid laden macrophages, plasma cells, neutrophils and lymphocytes [1, 2]. This kind of inflammation has been reported in kidney, gall bladder and anorectal area. Reported cases of xanthogranulomatous oophoritis are less than 15 in number. The most common age group for xanthogranulomatous inflammation is third decade; reported cases were 18 to 72 years [1–4].

The etiopathology of this condition is not certain, and the proposed causes are inflammation secondary to pelvic inflammatory disease, pelvic endometriosis, primary infertility, long-standing intrauterine devices, chemotherapy for breast cancer, in born lipid metabolism of macrophages and antibiotic use. The condition xanthogranulomatous inflammation was first described by Kunakemakorn in 1976. In female genital tract, most common site affected by xanthogranulomatous inflammation is endometrium followed by vagina, cervix, fallopian tube and ovary. Proposed organisms for this type of inflammation are bacteria like *Bacteroides fragilis*, *Escherichia coli*, *Staphylococcus aureus*, *Salmonella typhi* [2]. Few cases of rare association with uterine artery embolisation, secondary to talc use, diabetes, as a complication of typhoid are also been described [3]. Wather et al. proposed that malakoplakia in urinary system and xanthogranulomatous inflammation were identical chronic inflammatory diseases. Cytoplasmic concentric calcific bodies called Michaelis–Gutmann bodies are found in malakoplakia. Yener et al. in their research article suggested that xanthogranulomatous inflammation may have the predisposer or may follow chronic active follicular salpingitis. The histopathological features of xanthogranulomatous mass with follicle-like structures may be misdiagnosed as an adnexal neoplasm; hence, frozen section is mandatory to rule out malignancy even when the preoperative biopsies are conclusive of adnexal neoplasm with possible malignancies.

The presenting complaints are pain abdomen, mass per abdomen, fever, anaemia, loss of appetite and menstrual irregularities. On pelvic examination, adnexal mass is usually palpable with or without tenderness. On laboratory evaluation, features of chronic inflammation anaemia, increased leucocyte count and elevated erythrocyte sedimentation rate are found. On radiological findings, the features resemble malignant ovarian tumours. Lipid laden macrophages, foamy histiocytes, other chronic inflammatory cells like lymphocytes, plasma cells, occasional neutrophils with or without multinucleated or Touton giant

cells with no granuloma formation are the histological hall mark of xanthogranulomatous inflammation [2]. Immunohistochemical studies will show positivity for CD68 (foam cell positive), CD3 (T lymphocyte marker), CD 20 (B lymphocyte marker) and kappa and lambda (positive in polyclonal lymphocytes) [2]. In the present case report, first case presented with ovarian mass with involvement omentum, adhesion with adjacent bowel and pelvic structures, mimicking malignancy. The removal of tumour was also difficult due to the involvement of surrounding structures. Second case presented as tubo ovarian mass with fibroid uterus. The diagnosis came as a surprise in both the cases.

The management of xanthogranulomatous oophoritis is oophorectomy alone. The clinical imaging and intraoperative findings may lead to misdiagnosis as malignancy. Hence, arriving at the right diagnosis by both surgeon and pathologist is mandatory to avoid radical surgeries and adjuvant therapies. The pathologist needs to take multiple sections to rule out malignancies since few cases of malignancies in genitourinary tract with xanthogranulomatous inflammation in other part may coexist. Few malignancies may have inflammatory features; hence, it is a difficult to diagnose even in straightforward cases also with such a closely possible differential diagnosis.

Conclusion

Xanthogranulomatous oophoritis mimics malignancy posing diagnostic dilemmas. Clinching the right diagnosis is the key to right management. Knowledge of the condition and high level of suspicion by both surgeon and pathologist will help in the appropriate management. Treatment is mainly surgical.

Compliance with ethical standards

Conflict of interest None.

Research involving human participants and/or animals The study presented for publication does not involve any intervention as part of study and it is a case report.

Informed consent Informed consent from patients for publishing their clinical data has been taken prior to submission of the paper.

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