CASE REPORT

Xanthogranulomatous oophoritis mimicking malignancy: a diagnostic dilemma

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SUMMARY

Xanthogranulomatous inflammation is a chronic condition in which the affected organ is replaced by lipid-filled macrophages with plasma cells, lymphocytes and neutrophils. It is very rare in the ovaries. A case of xanthogranulomatous oophoritis in a premenopausal multiparous woman presenting with abnormal uterine bleeding and adnexal mass whose clinical and imaging findings suggested malignancy is reported.

BACKGROUND

Xanthogranulomatous inflammation is a non-neoplastic condition resulting in destruction of the affected organ. It usually involves the kidneys, urinary bladder, stomach, bones, testes, epididymis and gallbladder.¹ It is very rare in female genital organs and mainly involves endometrium.² Only a few cases with ovarian involvement have been reported to date.³ Although multiple predisposing factors such as pelvic inflammatory diseases, intra-uterine device use, leiomyoma uterus, endometriosis and inappropriate antibiotic intake are attributed to the development of xanthogranulomatous oophoritis, immunocompromised status leading on to chronic destruction of the affected organ, as in this case, has not been described so far.

CASE PRESENTATION

A 45-year-old woman, para 7 with six living children, presented with heavy vaginal bleeding for 20 days. She had a history of abdominal pain and menorrhagia for 1 year. She was addicted to tobacco and alcohol for more than 10 years. General examination revealed mild pallor and glossitis, pulse rate of 98 bpm and blood pressure of 150/100 mm Hg. Breast examination was normal. Abdominal examination revealed a firm, irregular, non-tender suprapubic mass arising from the pelvis. Speculum examination showed a 3×3 cm red polyp protruding through cervical os. On vaginal examination, the polyp was firm, smooth surfaced and bled on touch; its pedicle could not be reached. The uterus was 6–8 weeks in size, deviated to the left, with a right-sided 6×8 cm firm, non-tender adnexal mass, adherent to the uterus, with restricted mobility. Rectal examination was unremarkable.

INVESTIGATIONS

The patient's haemoglobin was 8.8 gm/dl% with dimorphic anaemia. Fasting and postprandial blood sugar was 192 mg/dl% and 220 mg/dl%, respectively. Liver and kidney function tests were within

normal limits. The patient's cancer antigen 125 (CA-125) level was 70.8 IU/mL. Ultrasonography of the pelvis revealed a complex, cystic mass of 8.5×7.3×7.3 cm in the right adnexa with internal septations and echoes, and the mass showed vascularity in its septae and periphery (figure 1). MRI revealed a multiseptated mass in the right adnexa with a mixed intensity matrix (figures 2 and 3).

Endometrial biopsy showed disordered proliferative endometrium on histopathology.

DIFFERENTIAL DIAGNOSIS

Ovarian carcinoma, endometrioma, tubo-ovarian mass.

TREATMENT

The patient was subjected to staging laparotomy in view of suspected ovarian malignancy after controlling hypertension and diabetes mellitus with antihypertensive and insulin injections, respectively.

Peroperatively, the uterus was deviated to the left, with dense adhesions between the ovaries, uterus, bowel and bladder, distorting the anatomy. There was a right-sided adnexal mass, cystic, around 8×8 cm within the broad ligament, which drained purulent fluid and the left ovary was enlarged, cystic and buried in adhesions. The uterus was 6–8 weeks in size, firm and regular, with a pedunculated polyp of 4×4 cm on cut section (figure 4). Lymph nodes were not palpable. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed.

Figure 1 Pelvic sonography showing a mixed echogenicity mass in right adnexa with multiple internal septations and echoes (left side). Doppler flow showing increased vascularity in septa and periphery of the mass (right side).

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Figure 2 Spin-echo T2-weighted image using a 1.5 Tesla MRI system shows a multiseptated cystic mass with mixed intensity, predominantly hyperintense matrix.

OUTCOME AND FOLLOW-UP

The patient was discharged on seventh postoperative day in satisfactory condition.

Histopathological examination of the ovarian mass showed dense inflammation comprising of sheets of foamy macrophages, lymphocytes, plasma cells and neutrophils. There was dense fibrosis with inflammatory cell infiltration, with no definite epithelial lining, consistent with xanthogranulomatous oophoritis (figures 5 and 6). The histopathology of the uterus showed a benign endometrial polyp.

DISCUSSION

Kunakemakorn *et al.*⁴ in 1976, was the first to describe xanthogranulomatous inflammation of serosa of the uterus, left

Figure 3 Spin-echo T1-weighted images reveal predominantly hypointense lesion with a few curvilinear areas of increased intensity.

Figure 4 Cut section of uterus with cervix—pedunculated polyp of 4×4 cm, with pedicle measuring 3.5×1.5 cm.

fallopian tube and ovary, in his report on inflammatory pseudotumours in the pelvis. The characteristic of this inflammation is infiltration of tissue by lipid laden histiocytes admixed with lymphocytes, plasma cells and polymorphonuclear leucocytes. The aetiology is unknown, but various associated risk factors are infection, inappropriate antibiotics therapy, use of intrauterine contraceptive device, abnormality in lipid metabolism, endometriosis, leiomyoma and recurrent pelvic inflammatory disease.⁵ Our patient had no history suggestive of pelvic inflammatory disease, endometriosis, intrauterine device or antibiotic intake, however, she was a chronic alcohol and tobacco consumer along with undiagnosed diabetes, and this must have led to the lower immunity and predisposed the body to infection. Ineffective immune response in combination with ischaemia, necrosis and haemorrhage, combined, might have resulted in formation of xanthoma cells along with lymphocytes, plasma cells, neutrophils and microabscesses, resulting in granuloma formation in this case.

These patients usually present with fever, abdominal mass, abdominal pain, menorrhagia, anaemia and tender adnexal mass. Our patient had all the above clinical presentations along with an enlarged uterus due to an endometrial polyp. Factors such as bleeding and obstruction may also initiate infection, tissue necrosis and release of cholesterol with other lipids, and phagocytosis by macrophages, as in this case, where the polyp

Figure 5 Photomicrograph showing infiltration of ovarian stroma by dense inflammatory infiltrate (×100 X, H&E).

Figure 6 Photomicrograph showing the characteristic foamy macrophages, neutrophils, lymphocytes and occasional plasma cells (x400 X, H&E).

in the uterine cavity caused obliteration and haemorrhage. These factors must have led to the development of chronic ovarian abscess. The inflammation of ovary may also involve neighbouring peritoneum leading to adhesion formation, as in this case.

Radiological features such as solid adnexal masses and involvement of adjacent structures due to the inflammation may mimic malignancy, as in this case. It is also difficult to diagnose this entity in CT scans as the appearance of a solid mass with altered signal intensity favours malignancy. MRI findings in xanthogranulomatous oophoritis cases are not very well described in the published literature. One may find multiple intramural nodules with thickened walls or well-defined septated cystic lesions that appear hyperintense on T1-weighted and T2-weighted MRIs, as reported in different case reports. These

lesions may also show mild peripheral and septal enhancement. The final diagnosis can only be made by histopathological examination. As in this case, xanthogranulomatous inflammation in other organs also has non-specific clinical findings, and is also difficult to diagnose clinically or on images and positron emission tomography or CT scan.

Learning points

- ▶ Xanthogranulomatous oophoritis must be kept as a differential diagnosis in a patient with enlarged complex cystic mass as the imaging modality is not diagnostic.
- ▶ Histopathological examination is the gold standard for the diagnosis of xanthogranulomatous oophoritis.
- ▶ Unnecessary and extensive surgery can be avoided by using the facility of frozen section histopathology in cases of dilemma.

Competing interests None declared.

Patient consent Obtained.

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