CASE REPORT

Xanthomatous oophoritis following uterine artery embolisation: successful conservative surgical management with favourable outcome

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SUMMARY

Xanthomatous oophoritis is a rare inflammatory condition of the ovaries. We are reporting a case of a 28-year-old woman, who had undergone uterine artery embolisation 4 years ago due to symptomatic focal adenomyosis. After 3 years of embolisation, the patient started having chronic pelvic pain, dysmenorrhoea and polymenorrhagia along with inability to conceive. Abdominal examination showed 16 weeks size mass arising from the pelvis. Ultrasound findings were suggestive of focal adenomyosis and bilateral tubo-ovarian masses. A CT scan report showed cystic enlargement of ovaries showing high-density fluid contents. MRI showed two well-defined, thick-walled, septated, cystic lesions appearing hyperintense on both T1-weighted (T1w) and T2w images with peripheral and septal enhancement. Bilateral abscess walls were excised and the healthy ovarian tissue was left behind during surgery. Histopathology of the cyst wall showed xanthomatous oophoritis. After the conservative surgery, she received three doses of goserelin. She conceived spontaneously thereafter and delivered a healthy term baby.

BACKGROUND

Xanthomatous oophoritis is a chronic, destructive and inflammatory condition of the ovaries caused due to infection with *Escherichia coli*, *Staphylococcus aureus* or actinomycosis and characterised by foamy, lipid-rich histiocytes, eosinophils, lymphocytes, macrophages and plasma cells. Fibrosis and vascular proliferation usually occurs and extensive tissue destruction usually follows. Many organs have been reported to be affected by xanthomatous inflammation like the kidneys, urinary bladder, stomach, bones, testis, epididymis and gall bladder.¹ Female genital organ involvement is rare and usually follows pelvic inflammatory disease or endometriosis.^{2–3} The predisposing factors are necrosis, haemorrhage and obstruction. Association of xanthomatous oophoritis with leiomyoma uterus, Crohn's disease, diverticulitis, reaction to gloves powder left during prior surgery and intrauterine device usage has been reported.^{3–6} We are reporting a 28-year-old patient of adenomyosis uteri, who had undergone uterine artery embolisation 4 years ago, and developed xanthomatous oophoritis. This case is reported due to rarity of this condition as there are 16 cases of ovarian xanthomatous inflammation reported until now to authors' knowledge.^{1–16} Uterine artery embolisation, as a

predisposing factor to this condition, has not been reported earlier. MRI findings of xanthomatous oophoritis are discussed in only one previous case report.⁷ Successful pregnancy outcome in these patients has also not been reported earlier.

CASE PRESENTATION

A 28-year-old nulliparous woman, who was married for 2 years presented to the gynaecology clinic with polymenorrhagia, dysmenorrhoea and dull aching and constant lower abdominal pain since 9 months. She also presented with inability to conceive and her preliminary investigations for infertility were within normal limits. She had a history of menorrhagia and was diagnosed as having focal adenomyosis for which she underwent bilateral uterine artery embolisation 4 years ago. Ultrasonography (USG) performed prior to embolisation revealed a focal adenomyosis of size 3.5×3 cm with normal bilateral ovaries. She did not respond to medical management. Embolisation was performed using gelfoam and a coil. She remained asymptomatic for 8 months after embolisation and started having pelvic pain thereafter and was managed by oral and parenteral analgesics prescribed by a local physician. After 3 years of embolisation she started having menorrhagia, severe dysmenorrhoea and constant dull-aching pelvic pain. Abdominal ultrasound scan revealed the uterus was enlarged to 7.6×5.8×4.2 cm with few ill-defined hypoechoic lesions suggestive of adenomyosis and bilateral tubo-ovarian masses of size 5.3×4.9 cm and 5.8×5.2 cm. A CT scan of abdomen and pelvis was performed on the advice of her local physician. The CT scan report showed an enlarged uterus with distorted outline due to an ill-margined mass in the posterior uterine wall suggestive of adenomyotic patch. Bilateral ovaries were enlarged with thick-walled cystic lesions showing high-density fluid contents likely to be endometriotic cysts (figure 1). No free fluid or any other abdominal pathology was noted on CT scan. She was put on some treatment by the private practitioner, but had only minor symptomatic relief. After 1 year of continuing this treatment, the patient presented to us as she was not relieved of symptoms.

General examination at presentation revealed moderate pallor. Abdominal examination revealed irregular, firm and tender mass corresponding to 16 weeks sized gravid uterus arising from the pelvis with restricted mobility. Pelvic examination

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Figure 1 A CT scan showing the enlarged uterus with distorted outline by an ill-margined mass in the posterior myometrium, likely focal adenomyosis. Enlarged ovaries showing thick-walled cystic contents with high-density fluid contents.

revealed 8–10 weeks size uterus and a large abdominal mass filling large area of the pelvis mainly on the right side.

INVESTIGATIONS

Routine blood and urine investigations were performed on presentation. Her total leucocyte count was 24 000/dL with 68% neutrophils, 30% lymphocytes and 2% monocytes. Erythrocyte sedimentation rate was 85 mm per first hour. Other routine blood and urine investigations were normal. Her cancer antigen 125 (CA-125) was 35.88 IU/mL. Transabdominal ultrasound revealed enlarged uterus with thickened myometrium and fluid collection in the uterine cavity. There were bilateral, large,

multiseptated, thick-walled cystic ovarian masses which were suggestive of endometriomas. MRI was performed to confirm the USG finding, which revealed uterine size of 10.7×6×6.6 cm and showed a 1.2×2 cm sized irregular area of altered signal pattern in posterior myometrium appearing hypointense on T1-weighted (T1w) and T2w images. Uterine cavity was distended with lobulated heterogeneous contents, appearing hypointense, with thin hyperintense periphery suggestive of blood clots. Two well-defined, thick-walled, septated, cystic lesions appearing hyperintense on both T1w and T2w images were noted in bilateral ovaries measuring 12×10.9 cm in size on the right side and 7.2×6.7 cm on the left side. The lesions showed peripheral and septal enhancement. Findings were suggestive of focal adenomyosis in the posterior uterine wall and bilateral endometriomas (figure 2).

DIFFERENTIAL DIAGNOSIS

The prime differential diagnosis in our patient was bilateral endometrioma associated with adenomyosis. The clinical features and imaging studies including MRI were suggestive of endometrioma. It was only after the surgery and histopathology report that we could make the final diagnosis. Other differentials were benign and malignant ovarian tumours. Tubo-ovarian masses due to chronic infective aetiology like tuberculosis or actinomycosis could also be the differentials.

TREATMENT

We planned for laparoscopy and ovarian cystectomy of bilateral endometriomas. After preoperative workup, we performed laparoscopy and found two large ovarian masses with dense adhesion to the bowel and the omentum. Since laparoscopic resection of such masses with such dense adhesions was beyond

Figure 2 MRI showing (A) T2-weighted (T2w) axial image, (B) T1w axial image, (C) postgadolinium image and (D) sagittal view. A 1.5T magnet system with spin echo (SE), turbo spin echo (TSE), short tau inversion recovery (STIR) and half-Fourier acquisition with single-shot turbo spin echo (HASTE) sequences were used. Enlarged uterus with an altered signal pattern in the posterior myometrium appearing hypointense on T1w and T2w images was seen. Two well-defined, septated, cystic lesions appearing hyperintense on T1w and T2w images are noted in bilateral ovaries. The lesions showing mild peripheral and septal enhancement.

our expertise, we proceeded for laparotomy. Operative findings revealed an enlarged right ovarian mass of 12×12 cm size and left ovarian mass of 10×8 cm size with dense adhesions all around the bowel, omentum, uterus and Pouch of Douglas (figure 3). Both fallopian tubes were stretched over the masses but they looked normal. The uterus was enlarged uniformly to 8 weeks size. While dissecting the adhesions of masses, accidental rupture of cyst occurred and foul smelling, purulent, yellowish-green coloured, thick in consistency, fluid drained out. The cysts were multiloculated with thick-walled septations. Bilateral abscess walls were excised and sent for histopathology. While excising the cyst wall, special care was taken not to injure the fallopian tubes. No endometriotic deposits were appreciated. A thorough lavage of peritoneal cavity was performed. The patient was administered injectable ciprofloxacin and metronidazole initially. After the pus culture and sensitivity report which showed the growth of *Klebsiella* species, we injected piperacillin and tazobactam for 1 week as per the sensitivity report. Her postoperative period was eventful and she remained afebrile. She was discharged on the 10th postoperative day. Histopathology report showed destruction of ovarian stromal tissue and invasion by inflammatory cells like foamy histiocytes (xanthoma cells), lymphocytes and plasma cells. Findings were suggestive of xanthomatous oophoritis (figure 4).

OUTCOME AND FOLLOW-UP

Owing to dense adhesions and infective aetiology, we were sceptical about the fertility of the patient. We gave her 3 monthly doses of goserelin 3.6 mg in the postoperative period. We counselled the patient regarding fertile period and asked her to try for conception after 3 months. To our surprise, after 6 months

Figure 4 A histopathological slide showing ovarian stromal infiltration by sheets of foamy histiocytes with admixed lymphocytes, plasma cells and eosinophils. Inset showing high-power view.

of surgery, the patient conceived spontaneously. The transvaginal ultrasound performed in first trimester revealed normally growing fetus and normal bilateral ovaries. She had an uneventful pregnancy and delivered a healthy baby vaginally at term eventually. She was asymptomatic for 4 months after delivery during her follow-up.

DISCUSSION

The pathogenesis of xanthomatous inflammation remains controversial. Untreated or undertreated infection, ischaemia, tissue necrosis, ineffective immune factors and phagocytosis or

Figure 3 The right-sided ovarian cyst of size 10×10 cm seen during laparotomy. Successful ovarian cystectomy could be performed owing to a comparatively well-formed pseudocapsule. The left-sided ovarian cyst was of size 12×12 cm. Adhesiolysis was performed to separate the cyst from the Pouch of Douglas, small bowel and omentum and the remnants of adhesions could be seen over the cyst wall. During cystectomy, accidental rupture occurred due to absence of a true cyst capsule with removal of foul smelling, and drainage of the thick, purulent fluid in the peritoneal cavity. The cyst wall held by a clamp could also partially be seen.

combination of these may lead to xanthoma cell production admixed with lymphocytes, plasma cells, neutrophils, microabscesses and granuloma formation. Xanthoma cells are histiocytes with vacuolated appearance due to lipid collection in cytoplasm. Predisposing factors for xanthomatous inflammation are pelvic inflammatory diseases, intrauterine device, endometriosis, inborn errors of lipid metabolism, inappropriate antibiotic therapy and radiotherapy.⁸⁻⁹ The bacterial agents responsible for such inflammation are usually *E coli*, *Proteus* and *S aureus*. Our patient did not give any history of pelvic inflammatory disease, endometriosis, intrauterine device or any other predisposing factor. Uterine artery embolisation has not previously been reported to predispose this condition but in our patient it could have been as it may lead to ischaemia. Other authors also believe that xanthomatous inflammation occurs either after ischaemia or chronic obstructive process leading to congestion and infection.¹⁰⁻¹¹

Female genital organs' involvement has been reported to be usually unilateral unlike in our patient in which bilateral xanthomatous oophoritis was seen following bilateral uterine artery embolisation.¹⁶ The age group of patient in previously reported cases varied from 21 to 73 years.¹⁻¹⁵ The clinical features as reported include menorrhagia, anaemia, anorexia, fever, abdominal pain and heaviness. Clinical examination may reveal tender pelvic masses and lump in the abdomen.¹⁶ Our patient presented with polymenorrhagia, chronic pelvic pain and lump in the abdomen. She also was unable to conceive, which could be due to dense adhesions formed in the pelvis or because of stretching of the tubes over the large ovarian masses. Infertility or subfertility is also associated with uterine artery embolisation. Subfertility has been reported in association with xanthomatous oophoritis in previous reports too.³

Erythrocyte sedimentation rate may be raised and there may be leucocytosis.¹⁶ Imaging studies may reveal adnexal masses. Imaging findings may reveal suspicion of ovarian neoplasia as the growth may be solid-cystic, tumor-like, nodular masses and the inflammation may even extend beyond the ovaries and involve adjacent organs and owing to its clinical and radiological resemblance to neoplastic lesions, it is sometimes denoted as 'pseudotumor'.⁸⁻⁹ USG may reveal a solid-cystic mass with septations and clear margins. A CT scan may reveal a well-defined solid mass with altered signal intensity, which may be falsely interpreted as ovarian malignancy.¹⁶ In our patient, the CT scan findings showed ovarian cystic enlargement with high-density fluid contents resembling endometriomas. MRI findings in xanthomatous oophoritis have been discussed in only one previous case report to authors' knowledge.⁷ As reported in previous case report, xanthomatous oophoritis was seen in axial T2w turbo spin echo MRI as high-signal-intensity mass with multiple low-signal-intensity internal septations.⁷ Irregularly thickened walls with hyperintense nodules clustered in the ovarian mass have been reported in this MRI. These MRI findings were similar to the MRI findings in our patient.

There are only few reported cases of xanthomatous oophoritis and as the pathology is destructive in nature, the treatment reported in most of the cases is salpingo-oophorectomy.^{2-3 10-16} Antibiotic therapy has been attempted by some but was unsuccessful in decreasing the size of ovarian mass.¹⁶ As our patient was a nullipara and had bilateral lesion, we wanted to preserve the ovaries. The ovarian mass had clear borders with a pseudocapsule. The normal appearing ovarian tissue was spared and involved ovarian pseudotumour was excised. In another similar report, a patient with subfertility had endometriosis and leiomyoma had to undergo hysterectomy with bilateral

salpingo-oophorectomy.³ Owing to extensive destruction, cut section of these lesions shows solid, fragile areas with haemorrhage and necrosis in most of the cases.^{1-8 10-16} In our case, the lesion was mainly cystic but the cyst wall showed areas of necrosis and haemorrhage.

Fertility issues associated with this condition has not been addressed due to rarity of this condition. The patient we reported was cohabitating for 2 years and had not conceived and was only able to conceive after the conservative surgery. As already stated, this destructive lesion mandates complete removal of involved ovaries most of the times. But in our patient, we could save part of ovary which later lead to successful pregnancy outcome.

Learning points

- ▶ Xanthomatous oophoritis is a chronic, destructive and inflammatory condition of the ovaries caused due to infection with various microbial agents and characterised by foamy, lipid-rich histiocytes, eosinophils, lymphocytes, macrophages and plasma cells.
- ▶ It may mimic benign or malignant ovarian tumours, endometriomas and tubercular tubo-ovarian masses. Diagnosis can be made only after histopathology.
- ▶ Clinical features may vary from menstrual disorders like polymenorrhea, menorrhagia and dysmenorrhoea, pelvic pain and abdominopelvic masses.
- ▶ MRI may reveal well-defined, thick-walled, septated, cystic lesions appearing hyperintense on both T1-weighted (T1w) and T2w images with peripheral and septal enhancement.
- ▶ Conservative surgery including removal of involved inflammatory mass and sparing the healthy appearing ovarian tissue can improve fertility in these patients.

Competing interests None.

Patient consent Obtained.

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REFERENCES

- 1 Gray Y, Libbey NP. Xanthomatous salpingitis and oophoritis. *Arch Pathol Lab Med* 2001;125:260-3.
- 2 Punia RS, Aggarwal R, Amanjit R. Xanthomatous oophoritis and salpingitis: late sequelae of inadequately treated staphylococcal PID. *Indian J Pathol Microbiol* 2003;46:80-1.
- 3 Abeyasundara PK, Padumadasa GS, Tissera WG, et al. Xanthomatous salpingitis and oophoritis associated with endometriosis and uterine leiomyoma presenting as intestinal obstruction. *J Obstet Gynaecol Res* 2012;38:1115-17.
- 4 Chouairy CJ, Hajal EA, Nehme YA. Xanthomatous oophoritis secondary to talcum powder. Case report and review of the literature. *J Med Liban* 2012;60:169-72.
- 5 Altanis S, Raweily E, Katesmark M. Xanthomatous endometritis and oophoritis secondary to diverticulitis. A rare cause of postmenopausal bleeding. *J Obstet Gynaecol* 2007;27:746-7.
- 6 Wlodarski FM, Trainer TD. Granulomatous oophoritis and salpingitis associated with Crohn's disease of the appendix. *Am J Obstet Gynecol* 1975;122:527-8.
- 7 Jung SE, Lee JM, Lee KY, et al. Xanthomatous oophoritis: MR imaging findings with pathologic correlation. *AJR Am J Roentgenol* 2002;178:749-51.
- 8 Kunakemakorn P, Ontai G, Balin H. Pelvic inflammatory pseudotumor: a case report. *Am J Obstet Gynecol* 1976;126:286-7.
- 9 McEntee GP, Goughlan M, Corrigan T, et al. Pelvic inflammatory pseudotumor: problems in clinical and histologic diagnosis: case report. *Br J Obstet Gynaecol* 1985;92:1067-9.
- 10 Pace EH, Voet RL, Melancon JT. Xanthomatous oophoritis: an inflammatory pseudotumor of the ovary. *Int J Gynecol Pathol* 1984;3:398-402.

- 11 Ladefoged C, Lorentzen M. Xanthomatous inflammation of the female genital tract. *Histopathology* 1988;13:541–51.
- 12 Minkowitz S, Friedman F, Henninger G. Xanthogranuloma of the ovary. *Arch Pathol* 1965;80:209–13.
- 13 Singh UR, Revathi G, Gita R. Xanthomatous oophoritis: an unusual complication of typhoid. *J Obstet Gynaecol* 1995;21:433–6.
- 14 Naik M, Madiwale C, Vaideeswar P. Xanthomatous oophoritis—a case report. *Indian J Pathol Microbiol* 1999;42:89–91.
- 15 Hemalatha AL, Rao S, Deepak KB, et al. Xanthomatous salpingo-oophoritis: a rare entity at an exceptional site. *Indian J Pathol Microbiol* 2007;50:607–9.
- 16 Zhang XS, Dong HY, Zhang LL, et al. Xanthomatous inflammation of the female genital tract: report of three cases. *J Cancer* 2012;3:100–6.

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