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Xanthogranulomatous endometritis with unilateral salpingo-oophoritis in a postmenopausal woman masquerading as a malignancy

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SUMMARY

Xanthogranulomatous endometritis (XGE) is a rare pathological entity which is characterised by sheets of foamy histiocytes and lymphoplasmacytic infiltrates. This condition can mimic endometrial carcinoma. We report a case, clinically suspected as carcinoma of the endometrium/ovary, which was diagnosed as XGE with left salpingo-oophoritis on histopathology.

BACKGROUND

Xanthogranulomatous endometritis (XGE) or histiocytic endometritis is a rare and chronic inflammatory condition of the endometrium.^{1–2} Xanthogranulomatous inflammation (XGI) is commonly seen in various organs such as kidneys, gall bladder, stomach, anorectal region, urinary bladder, testis, vagina, bone and salivary gland but involvement of female genital tract is very rare. XGI is characterised by an intense collection of foamy histiocytes, plasma cells, lymphocytes, a few polymorphonuclear cells, with or without the presence of multinucleated giant cells, which are seen surrounding and/or destroying the normal structures of the tissue affected.^{3–5} XGE can mimic endometrial carcinoma clinically and on imaging studies. It may lead to a fatal outcome if not treated in time.⁶ Till date, there have been very few cases reported in the world literature with varied presentations. Here, we report a case of XGE with salpingo-oophoritis presenting as pyometra causing a dilemma in diagnosis and management.

CASE PRESENTATION

A postmenopausal woman in her 70s, with past history of hypertension and type 2 diabetes, presented with pain in the suprapubic and left iliac regions with minimal white discharge per vaginum for 6 weeks. She didn't reveal any significant previous gynaecological history. On examination, the patient was afebrile and vitals were stable. General physical examination was normal and systemic examination revealed the scar of previous sterilisation. On abdominal examination she had mild tenderness in the suprapubic region but no mass was palpable. On speculum examination, parous external os with purulent yellowish discharge was noted which was non-foul smelling. Erosion was noted in the posterior lip of the cervix. On bimanual examination the uterus was retroverted, enlarged to

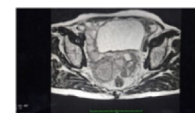


Figure 1 MRI showing hyperintense tubular fluid-filled lesion in the left adnexa, displacing the uterus right laterally and altered signal intensity fluid collection distending the endometrial cavity.

8–10 weeks size, and bilateral fornical tenderness was present.

INVESTIGATIONS

Blood investigations were done. The total and differential leucocyte counts were normal and the random blood sugar was 195 gm/dL with an glycosylated haemoglobin (HbA1C) level of 7.8. Pap smear showed dense sheets of neutrophils and numerous coccobacilli. Culture of high vaginal swab showed growth of *Klebsiella pneumonia* sensitive to various antibiotics. Ultrasonography of abdomen and pelvis showed hypoechoic fluid collection of 4.5×4.5 cm² size with internal echoes, in the upper uterine cavity suggestive of pyometra/serometra. A mixed echoic structure of 4.5×4 cm² abutting the left lateral body of uterus was also seen. MRI of the pelvis showed hyperintense, irregular, heterogeneous, predominantly solid, tubo-ovarian mass in the left adnexa and fluid collection of 4×3×3 cm³ distending the endometrial cavity suggestive of pyometra/haematometra (figure 1). She was evaluated for ovarian cancer and CA125 level was normal (24.8 IU/mL) giving an risk of malignancy index(RMI) Score of 74.4.

DIFFERENTIAL DIAGNOSIS

Infectious conditions like Gram-negative bacterial infections, fungal infections, malakoplakia and granulomatous infections like endometrial tuberculosis can present with vaginal discharge.

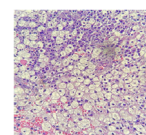


Figure 2 Histopathology showing sheets of foamy histiocytes replacing the endometrium.



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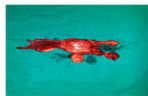


Figure 3 Gross specimen of uterus with bilateral tubes and left tubo-ovarian (arrow marked) mass with yellowish discoloration.

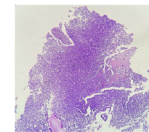


Figure 4 Histopathology showing sheets of foamy histiocytes replacing the endometrium.

Carcinoma of endometrium was considered due to the advanced age and presence of pyometra on ultrasound.

Carcinoma of endocervix was another differential diagnosis, as there was history of vaginal discharge with cervical erosion. However pap smear revealed absence of intraepithelial malignancy.

Carcinoma of ovary was another possibility due to the presence of tubo-ovarian mass on imaging, however RMI showed a low risk for malignancy.

TREATMENT

The patient was given antibiotics according to the culture and sensitivity report of the swab. Blood sugars were optimised. She underwent pyometra drainage along with endometrial biopsy. Histopathology of the endometrial sample revealed sheets of foamy histiocytes with lymphoplasmacytic infiltrates suggestive of XGE (figure 2). As there was involvement of the tube and ovary, she underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy. Intraoperatively the uterus appeared atrophic, the bilateral tubes were normal, the right ovary was atrophic and the left ovary showed a 5×4 cm² sized tubo-ovarian mass adherent to the underlying sigmoid colon and a 1×1 cm² hard yellowish nodule was seen in the left mesosalpinx. The cut surface of the tubo-ovarian mass was solid and yellowish in colour (figure 3). Histopathology of the hysterectomy specimen confirmed chronic XGE with intramural leiomyoma and chronic xanthogranulomatous salpingitis (figure 4). The nodule from the mesosalpinx showed chronic xanthogranulomatous oophoritis. The right ovary was unremarkable and there was no evidence of malignancy on histopathology.

OUTCOME AND FOLLOW-UP

The patient came for follow-up 6 weeks after surgery and was reassured about her histopathology report. She was asymptomatic and advised to maintain blood sugars within normal range with diet and medications.

DISCUSSION

XGE with salpingo-oophoritis is a rare disease characterised by chronic inflammation of the endometrium, tubes and ovary. The first case in the Indian population was reported by Barua *et al* in 1978.⁶ Not many cases have been reported so far in the Indian literature. The age of incidence ranges from 45 years to 88 years. The clinical features include lower abdominal pain, excessive vaginal discharge, pyometra and haematometra.^{7 8} One case of primary infertility with endometriosis and premature ovarian insufficiency has also been reported.^{9 10} Our case was a postmenopausal woman aged 70 years who also presented with lower abdominal pain and pyometra with *Klebsiella* infection. In a case report by Patel *et al*, ultrasound imaging showed unilateral tubo-ovarian mass of xanthogranulomatous salpingo-oophoritis which was similar to our case.¹¹ The pathogenesis of XGE is not well understood. Russack *et al* described that XGE involves a complex interaction of multiple factors, including cervical obstruction, inflammation and the presence of lipid, resulting in generation of free radicals and lipid

peroxidation.⁸ Other contributing features include necrosis and haemorrhage. Previous authors have also identified cervical stenosis as a risk factor for XGE. However it was not found in our case. Du *et al*, have reported diabetes mellitus as a possible risk factor for the development of XGI.⁷ Other possible risk factors include pelvic inflammatory disease, endometriosis, intrauterine contraceptive device and antibiotic treatment. Our case did not have any other risk factors apart from diabetes. Different bacteria like *Escherichia coli* and *Proteus vulgaris* have been recognised as contributing factors. *Klebsiella* subspecies was found in our case which is not reported so far as an aetiological organism. Histopathology is characterised by granulation tissue with foamy histiocytes, multinucleated giant cells, haemosiderin and calcium. Immunohistochemistry is found helpful in cases of diagnostic difficulty. As the diagnosis was evident in our case by histopathology, immunohistochemistry was not done.

Various granulomatous conditions such as tuberculosis and fungal infection are known to present as XGE. In our case all these conditions were excluded by histopathology. One-fourth of the cases reported so far were associated with malignancy. The non-specific symptoms and presentation makes the differentiation between XGE and malignancy difficult. Sampling of the entire endometrium is important to diagnose and eliminate the risk of endometrial carcinoma.² Early diagnosis and treatment of urogenital infection with appropriate antibiotics can prevent the development of chronic XGE. Surgery is sometimes the appropriate management especially when relapse occurs following antibiotic treatment. Surgical management was done in our case due to tubo-ovarian involvement and the suspicion

Patients perspective

I was told that there is a suspicion of malignancy of my womb and following surgery, it was a relief to know that i had a benign condition.

Learning points

- ▶ Xanthogranulomatous endometritis (XGE) is a pathological diagnosis and Gram-negative bacteria including *Klebsiella pneumonia* can cause XGE.
- ▶ Most of the cases of XGE may need hysterectomy and other granulomatous conditions like tuberculosis and fungal infections have to be ruled out. Early diagnosis and treatment with appropriate antibiotics may prevent the need for hysterectomy.
- ▶ Knowledge of this rare inflammatory condition is important for the gynaecologist and general physician so that the patient can be properly counselled.
- ▶ Frozen section can be done as there is a high chance of coexisting malignancy and if found positive one can proceed with pelvic lymphadenectomy.

of coexisting malignancy. Rarely death can occur secondary to systemic inflammation.⁵

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Case reports provide a valuable learning resource for the scientific community and can indicate areas of interest for future research. They should not be used in isolation to guide treatment choices or public health policy.

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