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 8–10 weeks size, and bilateral fornicaeal 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 pneumoniae 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, heterogenous, 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 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.

Figure 2 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.

OUTCOME AND FOLLOW-UP
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.

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. 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 ateration. The age of incidence ranges from 45 years to 88 years. The underlying sigmoid colon and a 1×1

of coexisting malignancy. Rarely death can occur secondary to systemic inflammation.²

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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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