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Case Report

Xanthogranulomatous endometritis: a benign mimicker of malignancy

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ABSTRACT

Xanthogranulomatous endometritis is a pathological entity which has an unusual characteristic of partial or complete replacement of the mucosa by granulation tissue with plenty of foamy histiocytes, sidero blasts and multinucleated giant cells. This condition clinically and radiologically mimics an endometrial carcinoma, which might create panic in patients as well as healthcare professionals. Hence it is important for Gynaecologists and pathologists to be aware of this condition so as to differentiate it from malignancy. Coexisting malignancy too is a possibility to be ruled out. We report case of a 69-year-old for whom we had high suspicion for malignancy based on clinical and radiological features, hence surgical oncologist opinion was obtained and total laparoscopic hysterectomy+bilateralsalpingo-oophorectomy and frozen section was performed-reported as inflammatory reaction. Hence, further oncological procedure (lymphadenectomy and omentectomy) was abandoned. The final Histopathology was reported as xanthogranulomatous endometritis. We are publishing this case report due to rarity(<30 cases reported worldwide) and its close resemblance to malignancy clinically and radiologically.

Keywords: Xanthogranulomatous endometritis, Malignancy, Granulation tissue

INTRODUCTION

Xanthogranulomatous inflammation is a well-established histological entity characterised by xanthogranuloma which is composed of foamy histiocytes, lymphocytes, plasma cells and variable number of foreign body giant cells. It is known to affect several organs, especially kidney, gallbladder, salivary glands and bones. Female genital tract (especially endometrium) involvement is rare.[1] <30 cases have been reported so far.[2] It bears a close resemblance to malignancy clinico-radiologically: hence posing a challenge for accurate diagnosis.

CASE REPORT

Herein, we report the case of a 69-year-old postmenopausal P2L2 with previous 2 normal vaginal deliveries who had complaints of 1 episode of postmenopausal bleeding and foul-smelling vaginal discharge-3 months ago, which lasted for 3 days. She presented to an outside hospital for the same. A forgotten Copper T was found and removed.

She denied history of fever, postcoital bleeding, discharge per vaginum, weight loss or loss of appetite.

There was no history of endometriosis/pelvic inflammatory disease in the past.

Her medical history was significant for type 2 diabetes mellitus, systemic hypertension and hypercholesterolemia for 20 years on regular medications.

She was initially evaluated at that hospital: Ultrasound whole abdomen showed thickened endometrium (7.9 mm) with irregular margins and tiny cystic spaces.

An MRI pelvis with contrast was performed which showed in homogenous lesion with ill-defined margin of size 28x15mm with cystic changes in endometrial cavity-hypointense in T1 W images and hypo to hyperintense in T2W images. It had features suggestive of malignant endometrium with inner myometrial invasion.

Hence an endometrial sampling was done and sent for histopathological examination which was reported to be a pyogenic abscess with bacterial colonies and inactive proliferative endometrium. No evidence of granuloma or malignancy was reported.

One month later, on her regular follow up visit, an ultrasound pelvis was done which showed endometrial thickness of 6.5 mm with endometrial cavity showing fluid (volume 15.24 cc) with internal echoes and echogenic content inside-suggestive of pyometra.

She was advised hysterectomy, for which the patient came to us for second opinion.

An ultrasound pelvis was done in our department-showed endometrial lining measuring 4 mm. Fluid with internal echoes measuring 5.4×4.0×3.8 cm (volume 43 cc) seen in the endometrial cavity (suggestive of pyometra).

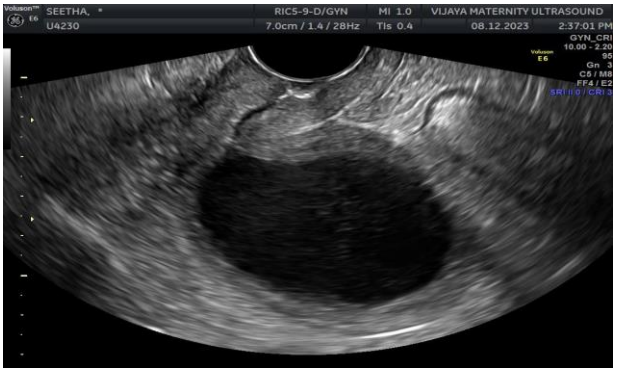

Figure 1: Ultrasound abdomen showing pyometra in this patient.

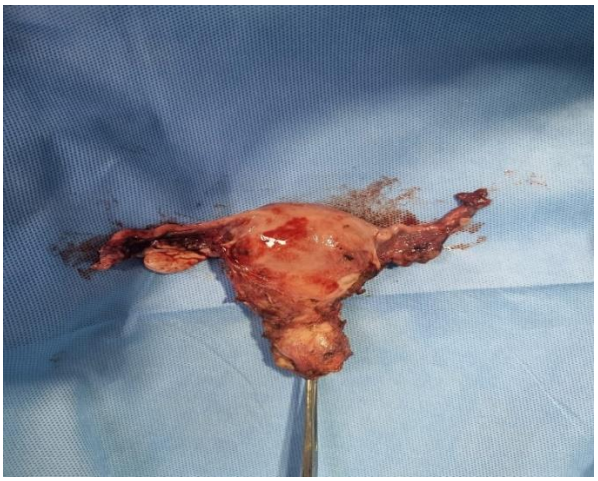

Figure 2: Anterior surface of specimen.

Based on her age, uncontrolled diabetes mellitus, pyometra and MRI report-there was high suspicion of malignancy but endometrial biopsy was reported as pyogenic abscess. With conflict between the diagnosis of malignancy versus infection, surgical oncologist opinion was obtained and patient was subsequently taken up for pyometra drainage+total laparoscopic hysterectomy+ frozen section with plan of oncological procedure(lymphadenectomy/omentectomy) if needed.

Intraoperatively, 100 ml of pus was drained-sent for culture and sensitivity. Uterus was found to be enlarged to 6-8 weeks size. Bilateral tubes and ovaries were normal. Total laparoscopic hysterectomy with bilateral salpingoophorectomy was done and specimen was sent for frozen section-reported to be inflammatory etiology. Hence further oncological procedure wasdeferred, avoiding morbidity to the patient. Patient was empirically started on IV Piperacillin-Tazobactam for 5 days. Post operative period was uneventful with no fever episodes. On POD 14, patient came for postoperative follow up-her vault was healthy with no vault cellulitis. There was no postoperative morbidity.

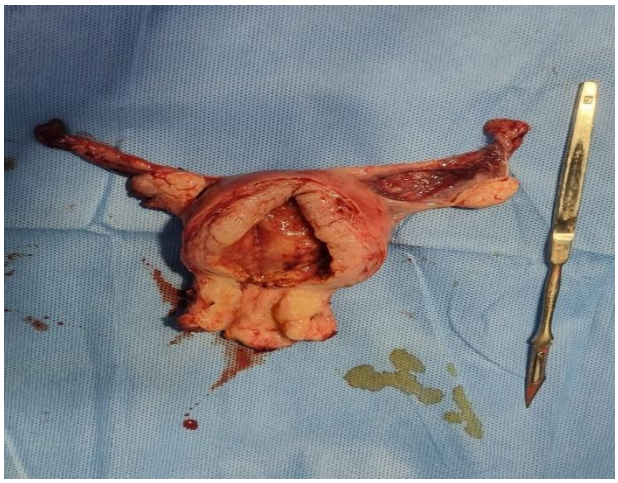

Figure 3: Cut section of specimen.

Pus culture and sensitivity showed no growth

Final histopathology report of hysterectomy specimen showed endometrial tissue with ulceration and dense mixed inflammatory cell infiltrate comprising of neutrophils, lymphocytes, plasma cells and numerous foamy macrophages.

The final diagnosis was consistent with pyometra with xanthogranulomatous endometritis.

Frozen section helped us to differentiate a benign condition from malignancy, hence we were able to decide against an oncological procedure-thereby avoiding morbidity, increased cost to the patient and prolonged hospital stay.

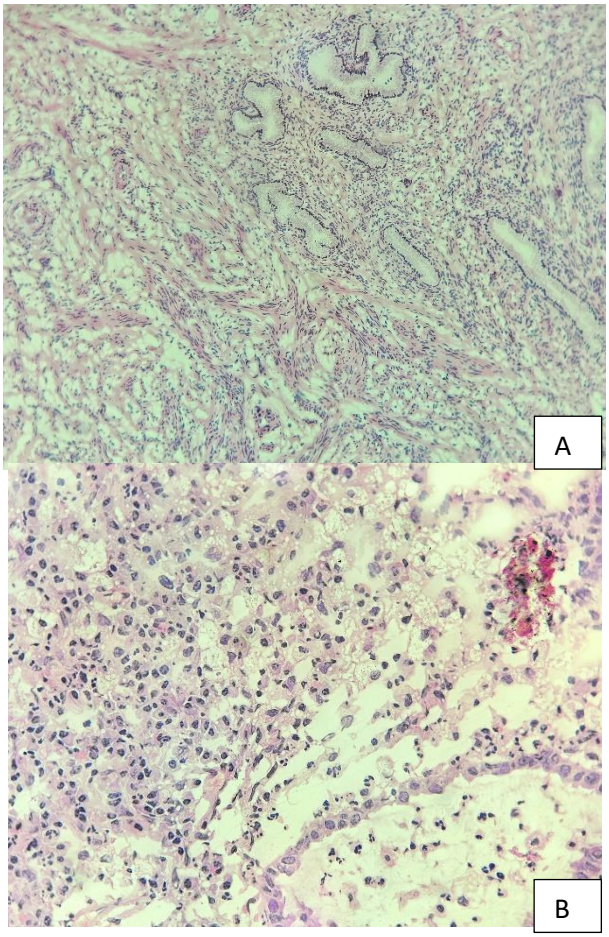

Figure 4 (A and B): Photomicrographs showing endometrial tissue with ulceration and dense mixed inflammatory cell infiltrate comprising of neutrophils, lymphocytes, plasma cells and numerous foamy macrophages.

DISCUSSION

Xanthogranulomatous endometritis (XE) is a rare pathological finding characterised by focal or total replacement of endometrial tissue by foamy histiocytes with variable amounts of associated multinucleated giant cells, other chronic inflammatory cells, necrosis, calcium, cholesterol clefts and hemosiderin. It is a benign masquerade of endometrial carcinoma. The irregular and necrotic appearance of XE may mimic carcinoma grossly, as reported by Ekici et al in 2007.

Synonyms used for XGE include histiocytic endometritis and pseudoxanthoma tous endometritis.

The first case of XGE was reported by Barua et al in 1978. <30 cases have been reported so far.

The age of onset is from 59 to 88 years, with a mean of 72 y.o. Bleeding, excessive vaginal discharge, and cervical stenosis with or without pyometra are the most common symptoms.[3] One case of a reproductive age woman (31 years) presenting with infertility too has been reported.[4]

Another uncommon presentation was of a 50-year-old with prolapsed uterus and endometrial calculi seen in cut surface of uterus.[5]

Russac and Lammers have reported 6 cases of XE, all associated with endometrial carcinoma that had been irradiated with external beam and/or intracavitary implants. Contributory factors such as necrosis, haemorrhage, obstruction, tumour bulk, or delay in treatment were identified.[6]

Mechanism of development of XE is still incompletely understood. The proposed mechanism involves a complex interaction of elements such as obstruction, inflammation and a lipid source, as well as generation of free radicals and lipid peroxidation, which are unique to the irradiated tissues.

Presence of cervical stenosis could lead to accumulation of endometrial secretions, infection, and tissue necrosis, and further release of cholesterol and other lipids which get phagocytozed by macrophages resulting in the formation of foam cells and XGE.

In a number of women, diabetes mellitus has been found to be a possible risk factor. Other risk factors include pelvic inflammatory disease, endometriosis, intrauterine contraceptive device and antibiotic treatment.[7]

Differential diagnosis includes endometrial carcinoma and Malakoplakia.

The infiltration of the myometrium by foamy macrophages might be misdiagnosed as clear cell carcinoma or sarcoma, but the cytological characteristic should lead to the XGE diagnosis. Immunohistochemistry proves to be helpful in the diagnosis of XGE in difficult cases. The presence of CD68 positive foamy histiocytes and a chronic lymphocytic infiltrate positive for CD3 and CD20 is in favour of an inflammatory process over carcinoma.

The absence of concentric calcific bodies (Michaelis-Gutmann bodies) and negative α1 antitrypsin staining of foamy histiocytes is helpful to exclude a malakoplakia.

Different bacteria, as Proteus vulgaris or Escherichia Coli, have been recognized as a contributing factor. However, there could be pyometra with no organism growth too, as described by Buckley and Fox.[8]

Recovery after antibiotic treatment or spontaneous resolution are the most frequent outcomes, but relapse is possible and radical surgery is then the appropriate treatment.

Lack of treatment seems to lead to a risk of systemic inflammation.

One case of death directly related to XGE has been reported in a 68 years old woman with a history of

abdominal pain. Death was caused by heart failure due to systemic inflammation, and pathological examination revealed XGE with transmural extension into the peritoneal cavity.[9]

The presence of XE does not rule out the possibility of a coexisting endometrial carcinoma. Because of this possibility, sampling of entire endometrium should be considered.

CONCLUSION

The pathogenesis of xanthogranulomatous endometritis is still incompletely understood. Age of onset is from 59-88 years with a mean age of 72 years. Bleeding, excessive vaginal discharge with or without pyometra are the most common symptoms with which a patient can present.

Xanthogranulomatous endometritis is a diagnosis of exclusion. We need to have a high suspicion for malignancy since this mimic's endometrial carcinoma closely and there can be coexistence of the two conditions too. At the same time, an Incorrect diagnosis can lead to unnecessary surgeries, hence increasing morbidity and cost to patients. Histopathological examination is the gold standard to establish the diagnosis.

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