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Case Report

Pathology Section

Xanthogranulomatous Endometritis: A Case Report with Rare Presentation

VAIBHAV BHIKA BARI[1], SURABHI SHASHANK RUIWALE[2]

ABSTRACT

Xanthogranulomatous Endometritis (XGE) is a rare, chronic inflammatory entity characterised by an inflammatory infiltrate of foamy cells, lymphocytes, plasma cells, and multinucleated giant cells. This condition mimics endometrial carcinoma both clinically and radiologically, thus requiring histopathology for diagnosis. Hereby, the authors present a case report of a rare presentation involving a 60-year-old postmenopausal female who presented with uterine prolapse. While XGE primarily presents as pyometra, in present case, it manifested alongside prolapse of the uterus. Only one other case in the literature has documented such a presentation. Histopathological examination confirmed the diagnosis of XGE. Given that this entity mimics endometrial carcinoma, awareness of its aetiology, clinical presentation, and pathogenesis is crucial for clinicians and pathologists.

Keywords: Postmenopausal, Prolapse, Pyometra

CASE REPORT

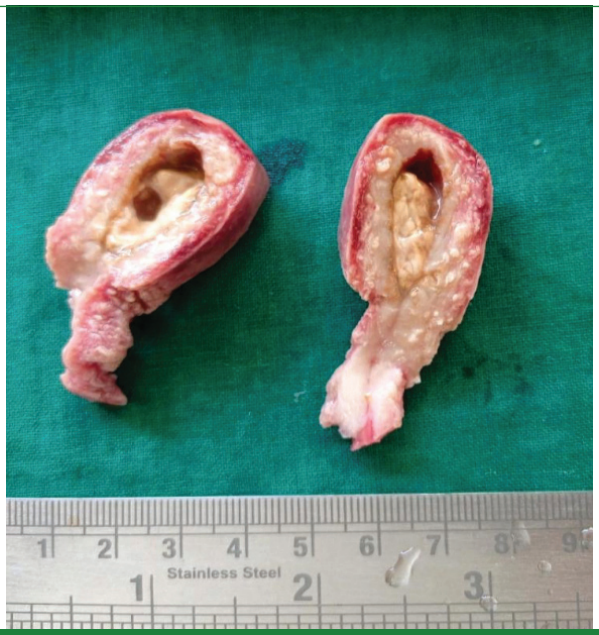
A 60-year-old postmenopausal female (P5L5A2) presented with complaints of something coming out of her vagina for six months, accompanied by back pain. There was no history of postmenopausal bleeding, discharge, weight loss, or loss of appetite. She had no other chronic medical conditions or surgeries. On abdominal examination, findings were normal.

During per speculum examination, she exhibited grade 3 uterovesical descent, cystocele, and grade 2 rectocele. Upon pervaginal examination, the uterus was found to be atrophic, and both the vagina and cervix appeared normal. The perineum was lax. Pap smear findings were consistent with the appearance of a prolapsed uterus. The clinical diagnosis was uterine prolapse, and subsequently, she underwent vaginal hysterectomy along with anterior and posterior repair.

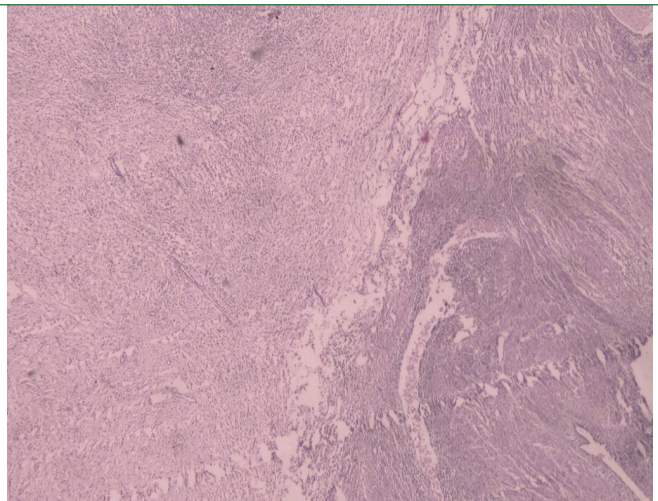
The Pathology Department received a uterus with cervix. The uterus measured 5×3.2×2.2 cm, and the cervix measured 3.4×3×1.2 cm. The external surface was congested but unremarkable. The cut surface showed yellow-white, thick necrotic material in the cavity, measuring 2×1×1 cm. The endometrial thickness was 0.1 cm, and the myometrial thickness was 0.4 cm.

There was no evidence of any endometrial growth or mass [Table/Fig-1]. Histopathological examination {Haemtoxylin and Eosin (H&E)} revealed that the endometrium was replaced by collections of foamy macrophages, lymphocytes, plasma cells, and hemosiderophages. The myometrium showed focal endometrial glands and stroma surrounded by smooth muscle. The glands were lined by cuboidal epithelium, with some glands being cystically dilated. Several blood vessels showed Monckeberg medial wall calcification.

Sections from the cervix exhibited the ectocervix with denudation. Focal ulceration and areas of mononuclear cell infiltration and congestion were observed [Table/Fig-2-4]. There was no evidence of granuloma, hyperplasia, or atypia. Since this condition was detected incidentally on histopathology and pyometra was not clinically suspected, no microbiological studies were performed. The final diagnosis was given as XGE. On postoperative follow-up, the patient recovered well without any complications.



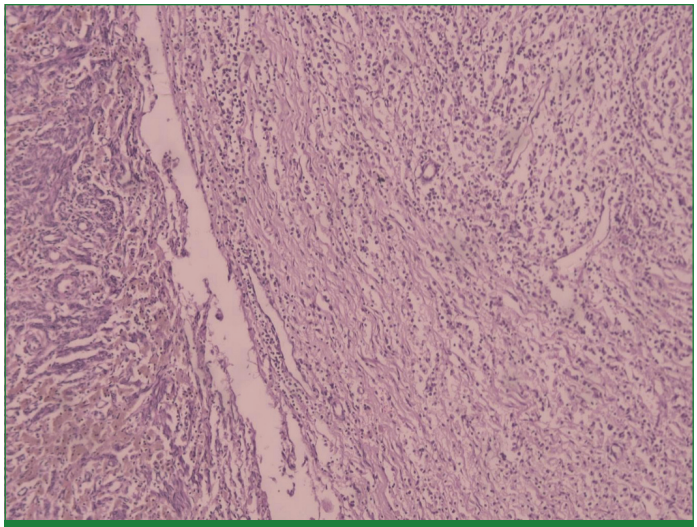
[Table/Fig-1]: Uterus cavity seen showing yellow-white thick necrotic material in cavity.



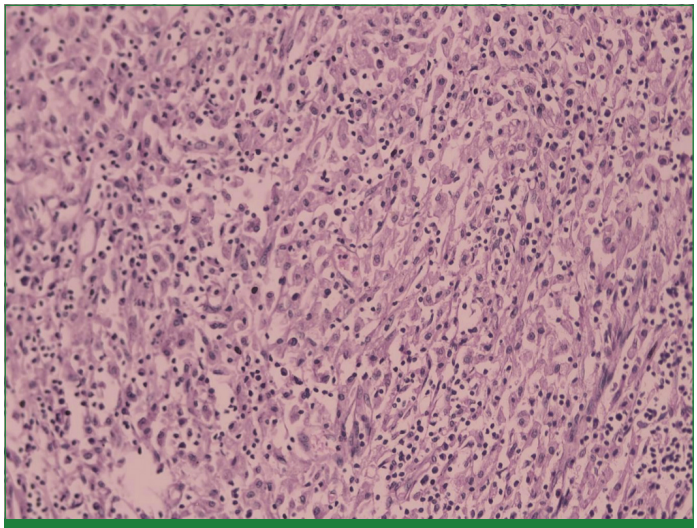
[Table/Fig-2]: Endometrial glands and stroma seen in myometrium on the left-side. Endometrium seen replaced by collections of foamy macrophages, lymphocytes, plasma cells and haemosiderophages (H&E, 4x).

DISCUSSION

Xanthogranulomatous Inflammation (XGI) is rare, frequently involving the kidneys and gallbladder, but is extremely rare in the uterus



[Table/Fig-3]: Collections of foamy macrophages, lymphocytes, plasma cells and hemosiderophages (H&E, 10x).



[Table/Fig-4]: Collections of foamy macrophages, lymphocytes, plasma cells and hemosiderophages (H&E, 20x).

[1,2]. The XGI of the female genital tract affects the endometrium, fallopian tubes, and ovaries [1]. It is a benign condition characterised by the replacement of endometrial tissue with foamy histiocytes, multinucleated giant cells, and other chronic inflammatory cells. It may also exhibit areas of necrosis, calcification, cholesterol clefts, and haemosiderin pigment [2].

The present condition is a mimicker of endometrial carcinoma. The irregular, necrotic material of XGI may resemble endometrial carcinoma on gross examination, as reported by Ekici ID et al., in 2007 [3]. This entity is also known as histiocytic endometritis and pseudoxanthomatous endometritis [2]. The first case of XGI was reported by Barua et al., in 1978. To date, fewer than 30 cases have been documented with this diagnosis [2].

The age of onset ranges from 59 to 88 years, with a mean age of 72 years. In the female genital tract, it primarily involves the endometrium, fallopian tubes, and ovaries, presenting as endometritis and salpingitis (tubo-ovarian abscess) [1]. Common symptoms include bleeding, excessive vaginal discharge, and cervical stenosis, with or without pyometra [2]. It may also present as a mass-like lesion in the pelvic cavity with infiltration of the surrounding tissue.

Kumar N et al., reported a case of XGI in a 50-year-old female who presented with a prolapsed uterus [4]. Such a rare clinical presentation, as seen in present case, has not been widely reported in the literature. A case report by Rani E et al., indicates that the patient presented with infertility, which can be another rare manifestation [5].

The pathogenesis of present condition is unclear [6]. Various causative factors include postmenopausal cervical stenosis or cervical carcinoma, tumour bulk or death of tumour cells following irradiation, necrosis, intrauterine haemorrhage, and atherosclerotic vessels [1]. These factors may lead to chronic inflammation and pyometra observed in this condition. Other possible risk factors include diabetes mellitus, pelvic inflammatory disease, endometriosis, intrauterine devices, and antibiotic treatment [2,7].

The cellular components, which include necrotic tumour cells, inflammatory cells, and red blood cells, serve as a good lipid source, providing an essential environment for the development of XGE [1]. In some cases, infections by bacteria such as *Proteus, Escherichia coli, Bacteroides fragilis*, and *Salmonella* have been reported; however, in most cases, these organisms are not identified [8]. A strong association with cervical stenosis has been observed, which could cause pyometra and subsequently lead to XGI [2]. This may have been a possibility in our case as well, contributing to the prolapse of the uterus.

The radiological findings of this entity typically present as a heterogeneous uterine collection indicative of pyometra or hematometra. In some instances, these findings may suggest either benign or malignant pathology, potentially showing invasion into the uterine wall and serosa [8]. The differential diagnosis include endometrial carcinoma and Malakoplakia [2]. XGE mimics endometrial carcinoma due to its non specific clinicoradiological features, making it a crucial differential diagnosis.

It is also important to note that the diagnosis of XGE does not necessarily rule out the presence of neoplasia. XGE has been observed alongside endometrial hyperplasia or carcinoma in some cases [1]. Furthermore, this condition has been associated with endometrial adenocarcinoma [6].

The XGE can be classified into two groups: one consisting of only XGE and the other comprising XGE along with endometrial carcinoma [5]. Therefore, extensive or, in some cases, complete sampling of the endometrium is advised to check for the presence of endometrial carcinoma [3]. Immunohistochemistry can be useful for diagnosis in challenging cases. The presence of CD68-positive foamy histiocytes and a chronic infiltrate positive for CD3 and CD20 points to an inflammatory process rather than carcinoma [2,6]. The absence of concentric calcific bodies (Michaelis-Gutmann bodies) and negative α1-antitrypsin staining of foamy histiocytes helps rule out Malakoplakia [2,6]. In the present case, no Michaelis-Gutmann bodies were observed, and Periodic Acid Schiff's (PAS) staining was negative, thereby ruling out Malakoplakia.

Some granulomatous conditions, such as tuberculosis and fungal infections, are known to present as XGE [7]. This was excluded on histopathology in present case.

The outcome for most cases is recovery after antibiotic treatment or spontaneous resolution; however, relapse is possible and may require radical surgery [2,6]. Lack of treatment can lead to an increased risk of systemic inflammation, resulting in peritonitis and sepsis [2,9]. This complication was noted in a case report by Na JM et al., from Korea [9].

CONCLUSION(S)

Since, XGE mimics endometrial carcinoma both clinically and pathologically, knowledge and awareness of this rare entity are crucial for Gynecologists, Radiologists, and Pathologists. Radiologically, when pyometra is suspected, XGE should be considered as a differential diagnosis. Additionally, there is a possibility of co-existence of endometrial carcinoma along with XGE; hence, it is recommended to sample the entire endometrium in cases of XGE.

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PARTICULARS OF CONTRIBUTORS:
1. Associate Professor, Department of Pathology, Rajiv Gandhi Medical College and Chhatrapati Shivaji Maharaj Hospital, Kalwa, Thane, Maharashtra, India.
2. Senior Resident, Department of Pathology, Rajiv Gandhi Medical College and Chhatrapati Shivaji Maharaj Hospital, Kalwa, Thane, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Surabhi Shashank Ruiwale,
84/903, Kalpavruksha, Vasant Vihar, Thanee-400610, Maharashtra, India.
E-mail: ruiwale.surabhi@gmail.com

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