Case Report

Xanthogranulomatous Endometritis: A Benign Uncommon Masquerader of Malignancy

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Abstract

Xanthogranulomatous endometritis is an uncommon benign lesion characterized by the destruction of endometrium and replacement by the sheets of foamy histiocytes, lymphocytes, plasma cells, multinucleated giant cells, fibrosis, calcification, and accompanying polymorphonuclear leukocytes. It is commonly mistaken for malignancy clinicoradiologically, and therefore, histopathological examination is of utmost importance. We report the case of a 61-year-old postmenopausal female who presented with pyometra, bulky uterus, and cervical stenosis, and histopathology revealed XGE.

Keywords: *Endometritis, histiocytes, pyometra, xanthogranulomatous*

Introduction

Xanthogranulomatous inflammation (XGI) is a rare, yet well-recognized histopathological entity defined by diffuse infiltration of the organs by foamy and lipid-laden histiocytes admixed with lymphocytes, plasma cells, and neutrophils. It is frequently reported in the gallbladder and kidney.[1] XGI in the female genital tract has been principally observed as endometritis and/or salpingitis (tubo-ovarian abscess).[2,3] Xanthogranulomatous endometritis (XGE) is a challenging mimicker of endometrial malignancy clinicoradiologically, thereby necessitating knowledge of this uncommon inflammatory lesion.

Case Report

A 61-year-old postmenopausal female presented with a complaint of lower abdomen pain for the past 6 months. She denied any history of fever/postcoital bleeding/discharge per vaginum/weight loss or loss of appetite. She had no history of endometriosis, pelvic inflammatory disease, or use of any intrauterine device. Her family history was noncontributory. On physical examination, her vitals were stable. She was normotensive (blood pressure: 126/84 mmHg) and pulse rate was 80/min and was afebrile. Per abdomen examination was essentially normal. On per speculum examination, cervix was flushed with vaginal vault and was poorly visualized. Per vaginal examination revealed synechiae and fibrosis at the vaginal vault. The cervix was hard on palpation and bled on touch and the uterus was bulky. Per rectal examination was performed, and rectal mucosa and pouch of Douglas were normal. Transvaginal ultrasonography showed the collection in endometrial cavity of size 4.3 cm × 3.1 cm with homogeneous internal echoes suggestive of pyometra/hematometra [Figure 1]. Endometrial thickness was increased, and it was irregular. Hematological and biochemical investigations were within the normal limits. In view of cervical stenosis and thickened endometrium with pyometra in an elderly female, a clinical possibility of cervical or endometrial carcinoma could not be ruled out. Cervical biopsy done revealed chronic cervicitis, and cervical cytology revealed atrophic smear. Drainage of pyometra and endometrial biopsy was attempted twice; however; it could not be performed because of cervical stenosis. In view of persisting symptoms and risk of spontaneous perforation of pyometra, the patient underwent panhysterectomy. Intraoperative examination showed small and atrophic cervix and pyometra in the uterine cavity. There was fibrosis at the vaginal vault. Bilateral adnexa were atrophic with

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How to cite this article: Malik V, Chatterjee D, Goel B, Takkar N. Xanthogranulomatous endometritis: A benign uncommon masquerader of malignancy. J Mid-life Health 2019;10:206-8.

Access this article online

Quick Response Code:

Website: www.jmidlifehealth.org

DOI: 10.4103/jmh.JMH_6_18

no evidence of inflammation. Peritoneal fluid cytology was unremarkable. Macroscopically, endometrial cavity was dilated and filled with pus. The endometrium was thickened and irregular and was lined with yellowish friable material [Figure 2], although no endometrial growth was seen. Histopathological examination showed destruction and replacement of the endometrium by the sheets of foamy histiocytes admixed with lymphocytes, plasma cells, few neutrophils, and occasional multinucleated giant cells [Figure 3]. Occasional tubular endometrial glands were preserved [Figure 4]. There was no evidence of endometrial hyperplasia or endometrial carcinoma or cervical carcinoma. Special histochemical stains such as periodic acid–Schiff (PAS) and Prussian blue stain were performed. No specific organism was detected on PAS. Prussian blue stain showed focal intracytoplasmic hemosiderin deposition in the macrophages. However, no intracytoplasmic inclusions were identified. The arteries in the parametrium showed atherosclerotic changes with marked intimal proliferation and luminal narrowing. Cervical tissue showed largely denuded epithelium and extensive fibrosis of the stroma. The final diagnosis was consistent with pyometra with XGE, secondary to cervical stenosis.

Discussion

XGI of the female genital tract affects the endometrium, fallopian tubes, or ovaries and presents as a mass-like lesion in the pelvic cavity with infiltration of the surrounding tissues.

XGE, also referred by some as histiocytic endometritis, is an unusual benign entity. The first case of XGE was discussed by Barua *et al*. in 1978.[4] Till date, <25 cases have been reported in the worldwide literature.[5] The age of onset ranges from 59 to 88 years, with a mean age of 72 years. The common presenting symptoms include excessive vaginal discharge and cervical stenosis with or without pyometra.[6]

The pathogenesis of XGE still remains debatable. The various causative factors implicated are chronic inflammation associated with pyometra due to

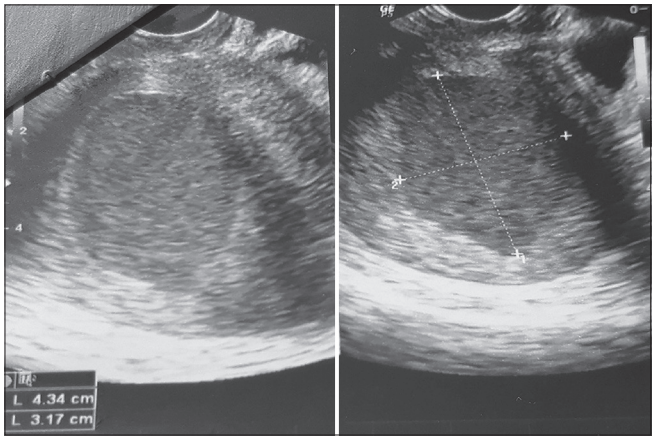

Figure 1: Transvaginal ultrasonography showing collection in the endometrial cavity, suggestive of pyometra/hematometra

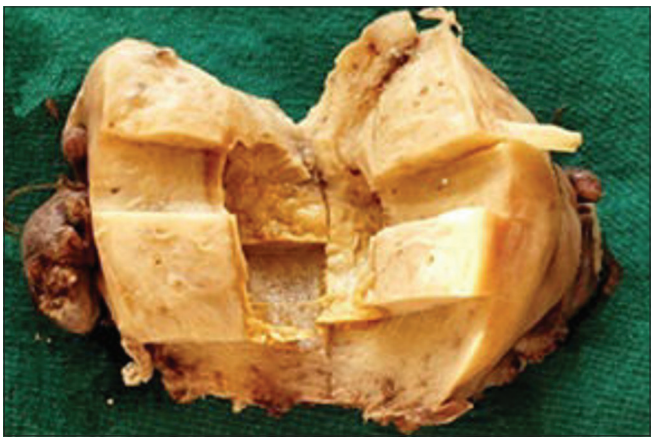

Figure 2: Dilated endometrial cavity and covered with yellowish friable material

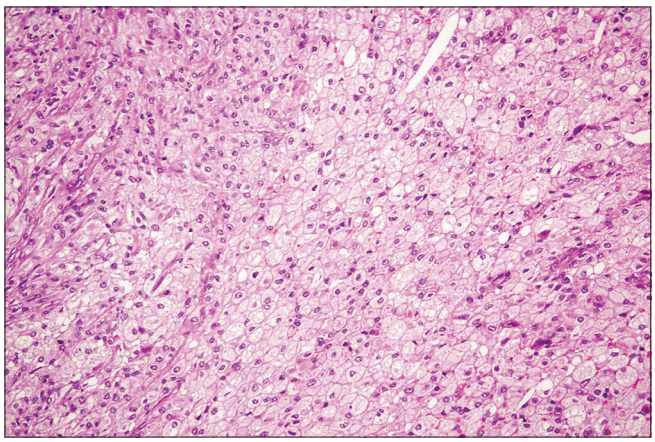

Figure 3: Sheets of foamy histiocytes replacing the endometrium (H and E, ×400)

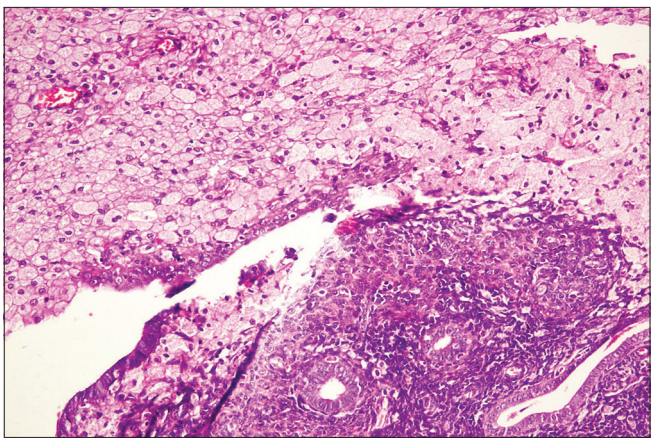

Figure 4: Destruction of the endometrium by the sheets of foamy histiocytes (H and E, ×400)

postmenopausal cervical stenosis or cervical carcinoma, tumor bulk or death of tumor cells following irradiation, necrosis, presence of intrauterine hemorrhage, and preexisting vascular compromise including atherosclerosis. The cellular component (necrotic tumor cells, inflammatory cells, and red blood cells) is a good lipid source and provides an essential medium for the development of XGE.[2,7] Our index case had postmenopausal cervical stenosis leading to obstruction, accumulation of endometrial secretions, infection and tissue necrosis, and further release of cholesterol and other lipids. These are then phagocytozed by the macrophages resulting in the formation of foam cells and further XGE. Certain bacteria, including *Proteus* spp. and *Escherichia coli,* have also been recognized by some, as the contributing factors.[4] However, very often, neither organism is identified.[8] The parametrial arteries, in this case, also showed severe atherosclerotic changes with marked luminal narrowing. The resultant ischemia might have also contributed in the development of XGE.

The foremost histological differential diagnosis of XGI is malakoplakia. The histopathological diagnosis of malakoplakia is based on the demonstration of intracellular and extracellular laminated inclusions, called calcospherites or Michaelis–Gutmann bodies, and special foamy histiocytes, called von Hansemann cells.[9] Our case had the absence of Michaelis–Gutmann bodies in histochemical PAS and Prussian blue staining, which almost excluded this diagnosis. Malignancy forms another important differential of this lesion. Clinicoradiological and gross irregular necrotic appearance of XGE can be easily mistaken for malignancy as was also reported by Doğan-Ekici *et al*. in 2007.[1] Radiological and clinical inability to differentiate the close differentials poses a challenge for clinicians and pathologists and highlights the importance of histopathology. However, the existence of XGE does not rule out the presence of carcinoma. Endometrial hyperplasia and endometrial and cervical carcinomas have been reported to coexist with this entity.[2,7] Therefore, exhaustive sampling should be done to rule out the concomitant presence of any neoplastic focus, as was done in this case.

Majority of these cases resolve spontaneously or after antibiotic treatment.[2] However, a few, if untreated, can result in adverse consequences by causing systemic inflammation.[10] Surgery thus forms the mainstay of the treatment.

Conclusion

XGE is a close benign differential of malignancy, and radiological and clinical examination alone may not be enough to narrow down the diagnosis. Histological examination is essential to establish the diagnosis and exclude the mimickers.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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