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RESEARCH ARTICLE

XANTHOGRANULOMATOUS ENDOMETRITIS – AN UNUSUAL CAUSE OF POST-MENOPAUSAL BLEEDING

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ABSTRACT

Xanthogranulomatous endometritis is a rare benign pathology mimicking endometrial carcinoma which shows focal or total replacement of the endometrial tissue by granulation tissue with the presence of foamy histiocytes, variable numbers of multinucleated giant cells, chronic inflammatory cells, necrotic material, siderophages, calcium, and hemosiderin.Many a times, infiltration by foamy macrophages in the myometrium can bemisdiagnosed as clear cell carcinoma or sarcoma, Immunohistochemistry can help in coming to a definitive diagnosis. We hereby report the case of 56 years the postmenopausal lady with foul-smelling discharge PV clinically suspected to have malignancy, but histopathologically turned as xanthogranulomatous granulomatous endometritis.

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INTRODUCTION

Xanthogranulomatous endometritis (XGE) is an unusual type of chronic inflammation mainly affecting the kidney and gall bladder. [1] In the female genital tract it can rarely present as endometritis and oophoritis. [2,3] Xanthogranulomatous endometritis (XGE) is a rare entity with only 3 cases reported in Indian literature to date. [4,5] Its clinical and radiological resemblance to endometrial cancer makes its diagnosis difficult. Therefore, a thorough histopathological examination is required. It has an exceptional pathological finding of foamy (lipid-laden) histiocytes mixed with lymphocytes, plasma cells and polymorphonuclear neutrophils. [2]

CASE REPORT

A 56-year-old postmenopausal lady presented to the gynaecological OPD with the chief complaint of persistent foul-smelling yellowish discharge per vaginum for 3 days. She did not report any fever, pelvic pain or bleeding per vaginum. She had no history of pelvic inflammatory disease, IUCD insertion or genital malignancy i.e. cervical or endometrial in the past. She had no medical co-morbidity. Her physical examination was inconspicuous.

A gynaecological examination revealed an enlarged uterus with an erosive cervix. There was active pus discharge from the cervical os for which pelvic USG was requested. On pelvic ultrasound, there was gross distension of the uterus with echogenic content suggestive of pyometra around 145cc in volume with an endometrial thickness of 3mm. Bilateral adnexa were normal. Subsequently, she underwent USG-guided pyometra drainage with fractional curettage and cervical biopsy. An intrauterine pus collection of around 200 ccs was drained. The culture of pyometra was sterile after 48 hours. HPE report showed endometrial stroma was infiltrated by abundant foamy histiocytes, siderophages, neutrophils, eosinophils and plasma cells. These features were consistent with xanthogranulomatous endometritis.No evidence of tuberculous granuloma or malignancy was seen. In TB PCR no mycobacteria could be detected. Cervical biopsy was reported as mild on chronic cervicitis. She was kept under close follow up and her post-operative pelvic ultrasound one month later revealed no collection in the uterus with an endometrial thickness of 3mm. To date, she is asymptomatic after a close follow-up of 6 months.

DISCUSSION

Xanthogranulomatous endometritis also known as histiocytic endometritis or pseudoxanthomatous endometritis. [4] The first case of

XGE was reported by Barua et al in 1978 [6] and about 30 cases have been reported worldwide whereas in Indian literature only 4 cases have been reported till date. [4,5] It occurs at around 59 to 88 years with a mean age of 72 years [7]as in our case.

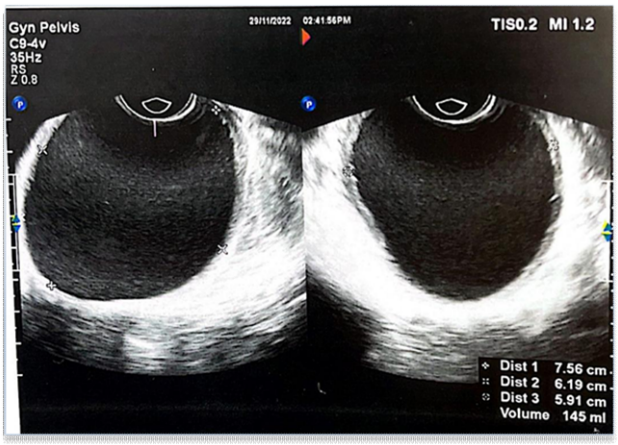

Figure 1. Pre-op USG s/o pyometra 7.5*6*5.9 cm – 145cc in volume

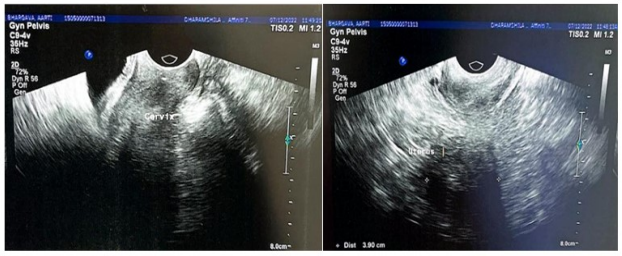

Figure 1. Post-op USG showing no collection in uterus

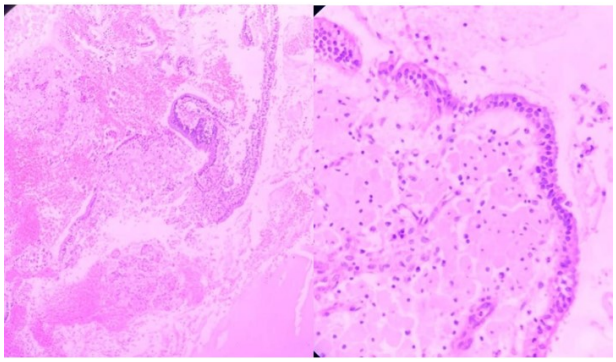

Figure 2. Replacement of the endometrium by granulation tissue with presence of foamy histiocytes, variable numbers of multinucleated giant cells, chronic inflammatory cells s/o xanthoogranulomatousendoometritis

XGE usually presents with pyometra or excessive vaginal discharge which was present in our case. Other presentations could be post-menopausal bleeding per vaginum, cervical stenosis, hematometra, anorexia, anaemia, fever, and pain in the upper abdomen. [8] An associated history of PID lasting for months to years is usually associated with it and antibiotic therapy seems ineffective in these cases. The risk factors commonly associated with this are diabetes mellitus, obesity, and previous history of radiation for cervical or endometrial cancer. [9]Although none was present in our patient. The exact cause of XGE is still not clearly understood and is debatable.Various causes have been put forward i.e., infection, endometriosis, intrauterine contraceptive device, and inborn error in lipid metabolism. [10] The theory of infection originated from various organisms like E.coli, pseudomonas, bacillus etc. isolated from the tissue by culture. [6] But the presence of a large amount of foam cells could not be justified by infection alone so it was suggested that it could be due to inappropriate use of antibiotics in the initial stages of infection which did not control bacterial proliferation leading to tissue necrosis thereby releasing cholesterol and other lipids which were later phagocytosed by macrophages and changed into foam cells. [10] The second theory suggests hyperlipidemia induced by lipid metabolic disorder. Foam cells are formed after phagocytes, phagocytose the lipid deposited in the affected region. XGE is characterized by focal or total replacement of the endometrium by granulation tissue with the presence of foamy histiocytes, variable numbers of multinucleated giant cells, chronic inflammatory cells, necrotic material, siderophages, calcium, and hemosiderin, as it was seen in our case. [11] The first and foremost histological differential diagnosis included in this case of XGE is malignancy. Malakoplakia is the second differential for XGE. Histologically the malakoplakia shows the presence of intracellular and extracellular laminated inclusions, called calcospherites or Michaelis–Gutmann bodies, and special foamy histiocytes, called von Hansemann cells, which were not identified histologically in our case. [12] Many times, infiltration by foamy macrophages in the myometrium can be misdiagnosed as clear cell carcinoma or sarcoma, Immunohistochemistry can help in coming to a definitive diagnosis. 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. [12] However, the presence of XGE does not exclude coexisting malignancy, [13] therefore, extensive sectioning and thorough microscopic examination of the whole endometrium is recommended. [14]In the 3 cases of XGE reported in Indian literature; one underwent a hysterectomy in view of a clinical picture resembling endometrial cancer. The other two cases were managed conservatively as in our case[4,5,10] The prognosis of xanthogranulomatous endometritis widely varies as some cases resolve spontaneously while others require antibiotic therapy. In cases of relapse, hysterectomy is the treatment of choice otherwise it can be managed conservatively.

CONCLUSION

Histological examination is essential to establish the diagnosis and exclude the mimickers. Awareness of this differential is essential for both gynaecologists and pathologists to prevent unnecessary hysterectomies for this benign condition resembling malignancy.

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