CASE REPORT

Xanthogranulomatous endometritis presenting as pyometra and mimicking carcinoma on imaging

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ABSTRACT

Xanthogranulomatous endometritis (XGE) is an uncommon but well-established histopathological entity seen affecting the kidney and gallbladder. Involvement of the endometrium is very rare, with only a few case reports in world literature till date. Histologically, it is characterized by the replacement of the endometrium by sheets of foamy histiocytes, plasma cells, lymphocytes, giant cells, and siderophages. We present a case of a 74-year-old female who presented with foul-smelling discharge and postmenopausal bleeding of a short duration. Clinical examination and imaging studies revealed a pyometra, cervical stenosis. A suspicion of carcinoma was raised. Since XGE may mimic an endometrial carcinoma clinically and pathologically, knowledge of this unusual and rare inflammatory pathology is important for both the gynecologists and the pathologists.

Key Words: Endometritis, histopathology, immunohistochemistry

INTRODUCTION

Xanthogranulomatous endometritis (XGE) or histiocytic endometritis (HE) is a rare and chronic inflammation of the endometrium.[1] XGE has been reported in various organs such as kidneys, gallbladder, stomach, anorectal region, urinary bladder, testis, vagina, bone, and salivary gland. However, involvement of female genital tract is very rare. XGE is characterized by an intense collection of foamy histiocytes, plasma cells, lymphocytes, 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.[2-4] The XGE inflammation may be so extensive so as to make an impression of endometrial carcinoma clinically and on imaging studies. It may lead to a fatal outcome in case if not treated in time.[5] Till date, there have been very few cases reported in the world literature. Here, we report another case presenting as pyometra and causing diagnostic misinterpretation of a carcinoma on imaging.

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CASE REPORT

A 72-year-old female, para 4 presented with complaints of foul-smelling yellowish-white discharge per vaginam for the past 4 days and postmenopausal bleeding for 1 day. She gave a history of hypertension for the past 1 year. There was no other significant personal or family history. On examination, the patient was moderately built and nourished. Per vaginal examination revealed a bulky uterus with clear bilateral fornices. There was no evidence of tenderness. On investigation, her routine biochemical and hematological parameters were within normal limits. Ultrasonography of the endometrium revealed a bulky uterus with a thickened endometrial lining measuring

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25 mm containing a pyometra. Magnetic resonance imaging of the abdomen confirmed the ultrasound findings and in addition revealed cervical stenosis [Figure 1]. A volume of 500 ml of pus was drained from the uterine cavity which was sterile on culture. In view of the clinical and radiological findings, a preoperative diagnosis of endometrial carcinoma was made. An endometrial biopsy was performed which revealed sheets of histiocytes having an abundant foamy cytoplasm, along with a dense mixed inflammatory infiltrate comprising plasma cells, lymphocytes, and neutrophils. Occasional hemosiderin-laden macrophages were also seen. No evidence of calcification was seen. Most of the endometrial glands were destroyed and an occasional remnant gland seen was atrophic [Figure 2a]. Special stains such as PAS, GMS, and Gram stain done did not reveal the presence of any organism. Prussian blue stain revealed intracytoplasmic hemosiderin accumulation in foamy histiocytes. Von Kossa stain did not reveal any calcium deposit. The foamy histiocytes were strongly positive for CD68 (for detection of monocytes and macrophages, ready to use, clone PGM1, mouse monoclonal, Dako Denmark.) [Figure 2b], and the presence of both CD3 (for detecting T-lymphocytes, ready to use, clone PS1, mouse monoclonal, Dako Denmark) and CD20 (for detection of B-lymphocytes, ready to use, clone L26, mouse monoclonal, Dako Denmark) positive lymphocytes suggested a polyclonal origin and thus an inflammatory process. Multiple sections studied did not reveal the presence of any malignancy. A final histopathological diagnosis of XGE was made.

DISCUSSION

Although XGE was first described by Barua *et al*. in 1978,[6] it is very uncommon with less than 25 reported cases in the world literature till date.[1]

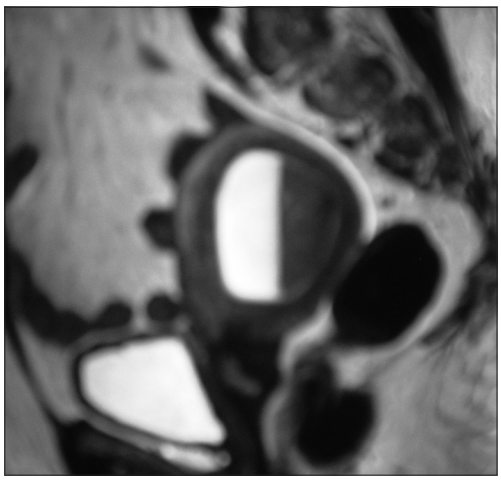

Figure 1: Magnetic resonance imaging of the abdomen revealed a bulky uterus with a thickened endometrial lining measuring 25 mm containing a pyometra. Cervical stenosis was seen in addition

Buckley and Fox coined the term "histiocytic endometritis" in 1980. However, the term "XGE" is preferred as it not only reflects on the inflammatory nature of the process but also unifies the similar morphologic findings seen in other organs.[7]

On histopathological examination, XGE is characterized by the presence of histiocytes with abundant cytoplasm which appears foamy and granular, variable numbers of multinucleated giant cells, chronic inflammatory cells, necrotic material, hemorrhage, calcium, and hemosiderin.

The pathogenesis of XGE appears to be chronic inflammation associated with pyometra due to postmenopausal cervical stenosis or cervical carcinoma. Russack and Lammers have reported six cases of XGE following irradiation of endometrial adenocarcinoma.[8] It is postulated that a complex interaction of elements such as obstruction, inflammation, generation of free radicals, and lipid peroxidation (as seen in irradiated tissue) is responsible for the development of this entity. Badhe *et al*. have reported a case of XGE in association with a malignant mixed Mullerian tumor of the uterus. The authors have proposed that rapidly growing tumors which show extensive areas of hemorrhage and necrosis that could itself lead to XGE in the absence of any irradiation.[9]

In our case, obstruction occurred due to cervical stenosis as there was no evidence of malignancy on histopathological examination.

Although some authors have proposed infection by *Escherichia coli* or *Proteus vulgaris* as a cause of XGE,[6] in our patient we could not isolate any organism either on culture

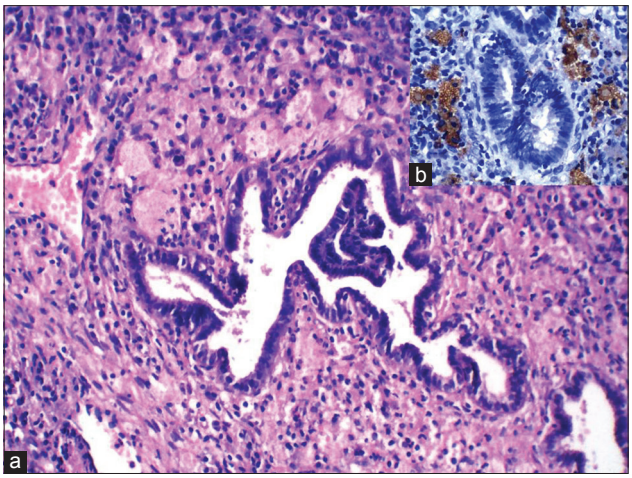

Figure 2: (a) Photomicrograph reveals sheets of histiocytes having an abundant foamy cytoplasm, along with a dense mixed inflammatory infiltrate comprising of plasma cells, lymphocytes, and neutrophils surrounding a distorted, atrophic endometrial gland. Most of the endometrial glands were destroyed and an occasional remnant gland seen was atrophic (H and E, ×400). (b) Inset: Foamy cells were positive for CD68 (×400)

or special stains. This is similar to the findings reported by other authors.[4,7]

The development of XGE may be influenced by various factors including tumor bulk, or death of tumor cells following irradiation, necrosis, presence of intrauterine hemorrhage, cervical stenosis, and preexisting vascular compromise including atherosclerosis. The necrotic tumor cells, inflammatory cells, and red blood cells are a good source of lipid and provide the necessary elements for the development of XGE.[8,10]

Immunohistochemistry proves 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 favors an inflammatory process over a carcinoma. The absence of concentric, calcific bodies (Michaelis–Gutmann bodies) and negative α1 antitrypsin staining of the sheets of foamy histiocytes are helpful clues to exclude a malakoplakia.

Not only the irregular, necrotic appearance of XGE may mimic a carcinoma on gross examination but also the sheets of histiocytes infiltrating the deeper tissue can mimic clear cell carcinomas or clear cell sarcomas and may pose a diagnostic challenge to a young pathologist.

Awareness of this entity is important for both the gynecologist and the pathologist as XGE can mimic malignancy on clinical examination and imaging studies. The presence of XGE does not rule out the coexistence of a carcinoma, which should be diligently searched for and requires extensive sampling of tissues to rule out any focus of malignancy.

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Conflicts of interest

There are no conflicts of interest.

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