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Research Letter

Xanthogranulomatous inflammation of myometrium with uterine perforation

Chi-Yuan Liao [a,*], Cheng-Hui Chiu [b], Fuh-Jinn Luo [c]

[a] *Department of Obstetrics and Gynecology, Mennonite Christian Hospital, Hualien, Taiwan*
[b] *Department of Radiology, Mennonite Christian Hospital, Hualien, Taiwan*
[c] *Department of Pathology, Mennonite Christian Hospital, Hualien, Taiwan*

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Xanthogranulomatous inflammation of the female genital tract is unusual and is essentially limited to the endometrium [1,2] and the fallopian tubes [3–6]. We report a rare case of xanthogranulomatous inflammation of the myometrium. This case is of interest because the xanthogranulomatous inflammation destroyed the myometrium and further extended to the pelvis with uterine perforation. The findings were first seen on ultrasonographic images and computed tomography (CT) with pathological confirmation after a surgical operation.

A 66-year-old married female, gravid 2, parity 2, with a history of poorly controlled diabetes, presented to the emergency room with complaints of lower abdominal pain, flank pain, and an intermittent fever of 1 week duration. She was febrile with a temperature of 38.2 °C.

Physical examination showed a mildly enlarged uterus with tenderness on the right adnexa. Ultrasonography revealed an enlarged uterus with pyometra, suspected uterine perforation, and a right tuboovarian abscess (Fig. 1B). Abdominal CT revealed pyometra and a pelvic abscess with a suspected uterine wall defect (Fig. 1A). The laboratory data showed the following details—white blood cell count, 23.63 THSD/μL; neutrophil, 86.6%; C-reactive protein, 18.30 mg/dL; and glucose spot, 536 mg/dL. The patient was admitted and received an antibiotic, Cefmetazole (Daiichi Sankyo, Tokyo, Japan) 2 gm IV Stat, followed by 1 gm IV Q6H. She then underwent an endometrial aspiration and Foley catheter drainage for her pyometra, which was acutely inflamed with numerous polymorphonuclear leukocytes and necrotic debris. Accordingly, she received a laparoscopic surgery. A thick adhesion mass about 5 cm in diameter on the right adnexa—affecting the sigmoid colon, omentum, right tube, and ovary—was found. Pus was found, and a culture was conducted. Adhesion lysis was performed, and a hole approximately 2.5 cm in diameter was observed on the right uterine fundus. A laparoscopy-assisted vaginal hysterectomy was performed smoothly. The culture reports from the blood, pyometra, and right adnexa were all negative. The pathological report of the uterus revealed localized xanthogranulomatous inflammation at the right uterine fundus with perforation (Fig. 2A). Postoperative antibiotic with Cefmetazole 1 gm IV Q6H was administered. Drainage using the Jackson–Pratt drain was carried out. The patient recovered without incident.

Xanthogranulomatous inflammation is rare. It mainly involves the kidneys or the gall bladder [1,4–7]. Primary xanthogranulomatous inflammation that involves the female genital organ has been reported in only a few cases, and it usually affects the endometrium or fallopian tubes. However, to date no case of myometrium involvement with uterine perforation has been reported. Xanthogranulomatous inflammation is a form of chronic inflammation that is destructive to the affected tissue, as in the case reported here, in which the myometrium was replaced with lipid-laden, foamy macrophages (xanthoma cells) and polymorphonuclear leukocytes, lymphoplasma cells (Figs. 2A and B). The etiology of the disease has not been fully elucidated. The proposed causes include occlusion of the cervical canal, hemorrhage, and infection [7,8]. Multiple factors may be responsible. In the case considered herein, cervix obstruction and infection with tissue necrosis occurred, followed by the release of cholesterol and other lipids. Therefore, phagocytosis by the macrophage may explain the formation of foam cells that is associated with xanthogranulomatous inflammation. In some cases, bacteria including *Proteus mirabilis* or *Escherichia coli* have been identified [9]. However, very often, neither organism [10] is identified, as in our case.

* Corresponding author. Department of Obstetrics and Gynecology, Mennonite Christian Hospital, Hualien, Number 44, Minchuan Road, Hualien City 970, Taiwan.

E-mail address: mchliaochiyuan@yahoo.com.tw (C.-Y. Liao).

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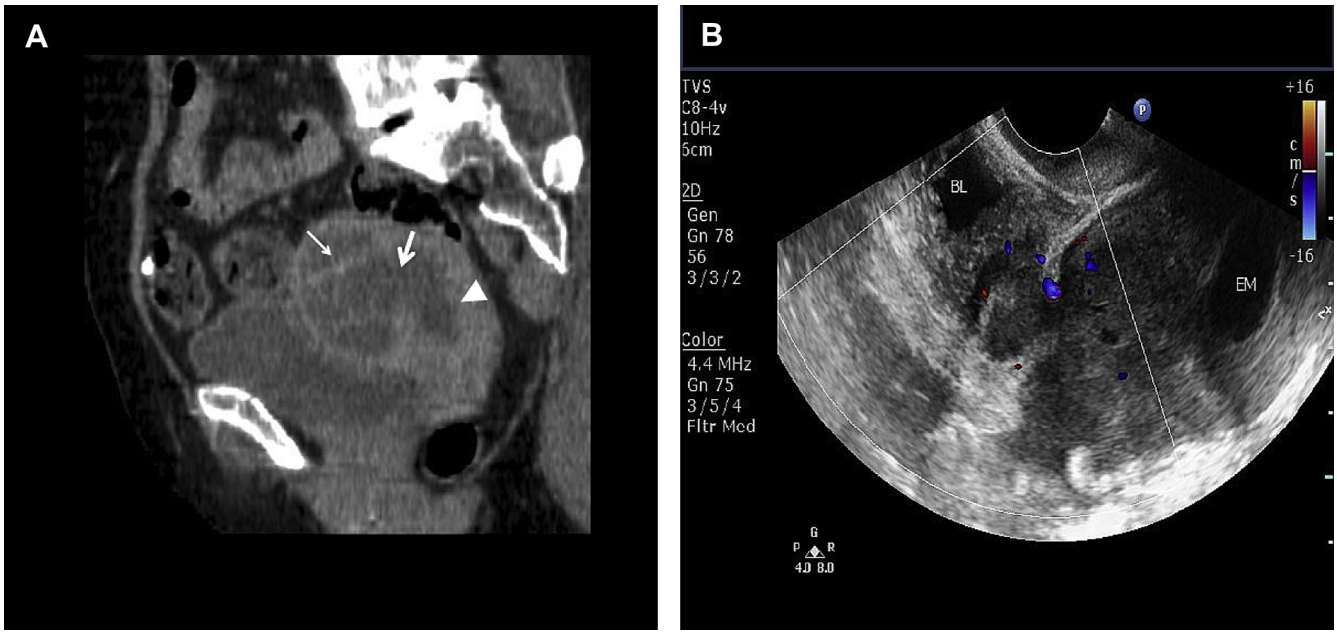

Fig. 1. (A) Oblique-sagittal reformation imaging showed dilated uterine cavity (arrowhead). Presence of a defect of myometrium at right anterior aspect (bold arrow) and abscess formation (arrow). (B) Ultrasonography revealed a perforation hole, about 2.5 cm in diameter, that was enclosed with an echogenic, septate mass 4 cm × 2.5 cm in diameter on the fundal region.

In our case, the histological involvement of xanthogranulomatous inflammation is present only in the myometrium with transmural extension to the peritoneal cavity. However, the endometrium and fallopian tubes were simply present with acute and chronic inflammation (Fig. 2A). This characteristic, pathological phenomenon associated with uterine perforation is similar to the formation of fistula in xanthomatous pyelonephritis and xanthomatous cholecystitis. Most cases of xanthogranulomatous endometritis resolve spontaneously or after antibiotic treatment [7,8,11]. In the case presented here,

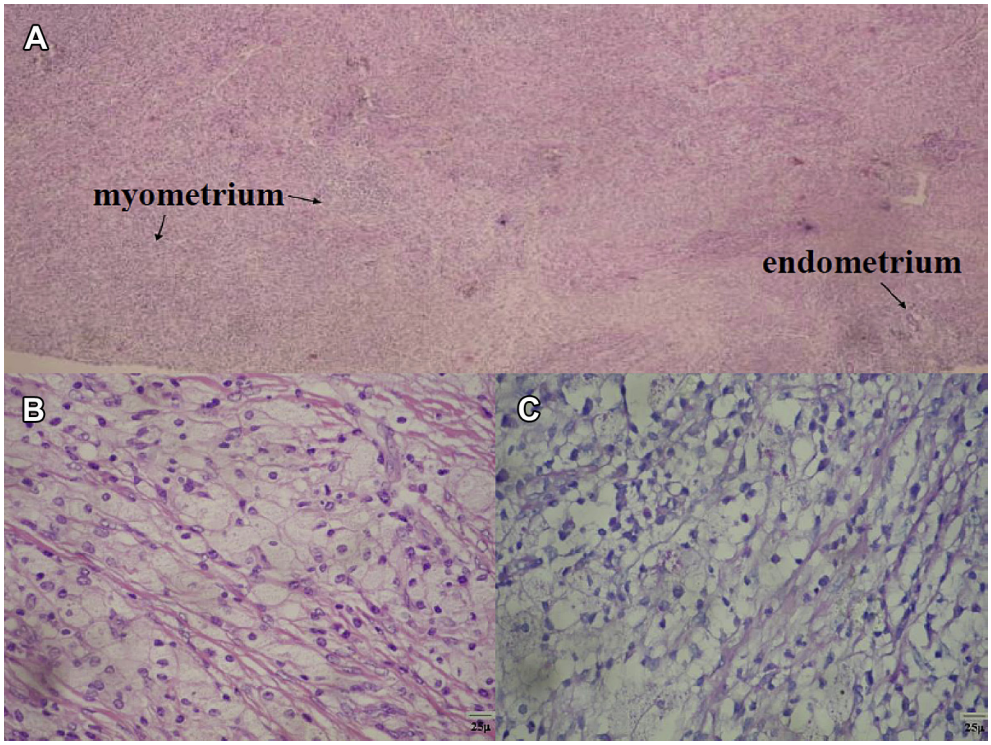

Fig. 2. (A) Xanthogranulomatous inflammation of the uterus. The myometrium displays xanthogranulomatous inflammation and overlying endometrium with usual inflammation. (hematoxylin–eosin, original magnification, ×40). (B) High magnification reveals abundant foamy histiocytes, admixture of lymphoplasma cells and neutrophils (hematoxylin–eosin; original magnification, ×400). (C) Periodic acid Schiff staining reveals no Michaelis–Gutmann body (original magnification ×400).

xanthogranulomatous inflammation destroyed the myometrium and extended to the pelvis with uterine perforation, as revealed by the ultrasonographic images and CT scan, which is an exceptional occurrence. Whether the patient's poorly controlled diabetes is a contributing factor is not known.

The most important differential diagnosis of xanthogranomatous inflammation is malakoplakia. The histopathological diagnosis of malakoplakia depends on the presence of intracellular and extracellular laminated inclusions, the so-called calcispherites or Michaelis–Gutmann bodies, and special foamy histiocytes, which are called von Hansemann cells [12]. In the case reported here, the lack of a Michaelis–Gutmann body in histochemical periodic acid Schiff staining almost excludes this diagnosis (Fig. 2C).

In conclusion, a rare case xanthomatous inflammation of the myometrium with uterine perforation associated with female genital organ infection is presented. A poorly controlled diabetes with a very high level of spot sugar (536 mg/dL) was noted. Whether it was a causal factor is uncertain. To the best of our knowledge, this is the first report of a xanthomatous inflammation of the myometrium with uterine perforation in a patient with poorly controlled diabetes. Surgeons will recognize the possibility of xanthomatous inflammation in pelvic inflammatory disease with pyometra, especially in patients with uncontrolled diabetes.

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