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Xanthogranulomatous Inflammation of Myometrium Causing Pelvic Extension: Report of Two Cases

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Abstract

Background: Xanthogranulomatous inflammation of the female reproductive organs is a rare chronic inflammation. In most reported cases, the lesion was limited to the endometrium and fallopian tubes. Here, we report two cases of xanthogranulomatous inflammation of the myometrium with a history of endometrial biopsy. Case Reports: In two cases, myometrial xanthogranulomatous inflammation destroyed the myometrium. This inflammation developed into surrounding pelvic organs, resulting in uterine perforation. Conclusion: When inflammatory lesions are found after intrauterine manipulation, the possibility of developing xanthogranulomatous inflammation should be considered. If antibiotics are ineffective, prompt surgical treatment is necessary.

Keywords

Xanthogranulomatous Inflammation, Myometrium, Klebsiella Pneumoniae, Surgical Intervention, Endometrial Biopsy

1. Introduction

Xanthogranulomatous inflammation is an uncommon type of chronic inflammation that can destroy normal tissue in affected organs. It primarily affects the kidneys, followed by the gallbladder, stomach, anal region, bones, bladder, testes and epididymis [1] [2]. Histopathologically, the diagnosis of xanthogranulomatous inflammation is made by marked proliferative fibrosis, destruction of the parenchyma, and infiltration of foam histiocytes mixed with other inflammatory cells. We report two rare cases of xanthogranulomatous inflammation of the

myometrium with a history of endometrial biopsy, in which the inflammation histopathologically destroyed the myometrium without invading the endometrium and further developed into the surrounding organs, resulting in uterine perforation.

2. Case Reports

Case 1

A 60-year-old (gravida 6, para 1) post-menopausal woman presented to our hospital with complaints of lower abdominal pain, vaginal discharge and an intermittent fever of three days' duration. She had undergone endometrial cytology uneventfully due to slight uterine bleeding and vaginal discharge two weeks before. Her past and family history was unremarkable. Magnetic resonance imaging (MRI) revealed an irregularly shaped mass, 6 cm in diameter, which was localized in the posterior wall of the uterus. On T1-weighted contrast-enhanced MRI, adhesions to the sigmoid colon and rectum were suspected (**Figure 1A**). The laboratory data showed the following details: white blood cell count, 14.400/μl; neutrophil, 83.3%; C-reactive protein, 7.88 mg/dl. Based on these findings, abscess formation in the uterus was suspected. The patient was admitted and received an intravenous antibiotic therapy but failed to respond. Accordingly, she underwent a laparotomy. The uterus was enlarged due to the myometrial abscess, and dense adhesions existed around the posterior wall of the uterus and sigmoid colon (**Figure 1B**). Adhesion lysis was performed and perforation approximately 3 cm in diameter was observed on the fundus. Total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed. There was neither inflammation nor enlargement in the bilateral adnexae. Klebsiella pneumoniae was detected in endometrial cavity. The postoperative course was uneventful.

Macroscopically, an abscess, 6 cm in diameter, was present in the posterior myometrium (**Figure 1C**). The cut surface of the abscess was purulent with hemorrhage, necrosis, and cystic degeneration. Microscopically, marked infiltration of foamy histiocytes with clear lipid-containing cytoplasm, together with abundant lymphocytes and plasma cells, was observed in the posterior myometrium and perimetrium (**Figure 1D**). The endometrium and fallopian tubes were simply present with acute and chronic inflammation. These findings were consistent with XGI of the uterine corpus.

Case 2

An-85-year-old (gravida 3, para 2) woman presented with lower abdominal discomfort, intermittent fever and leucorrhea on admission. She had undergone endometrial cytology without difficulties for cancer screening two months before due to vaginal discharge. Her past and family history was unremarkable. Physical examination showed a mildly enlarged uterus with marked tenderness. MRI revealed an irregularly shaped mass, 8 cm in diameter, which was localized in the posterior wall of the uterus with suspected uterine wall defect (**Figure 2A**). The white blood cell count and C-reactive protein were elevated to 24,700/μl

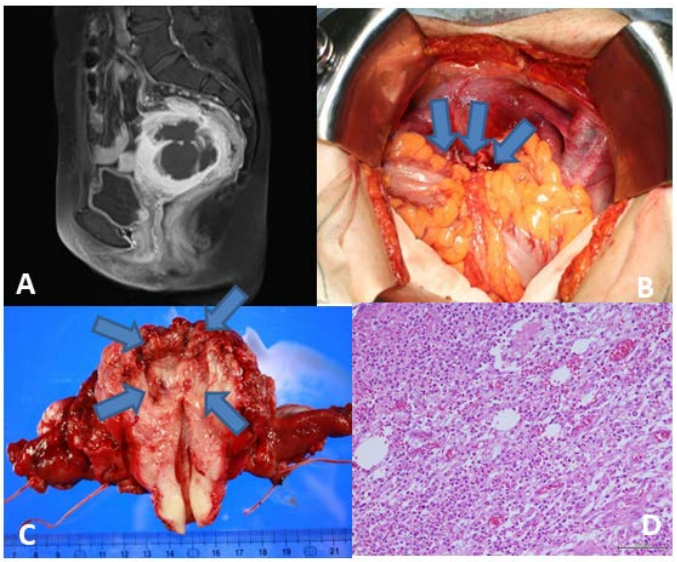

Figure 1. Case 1 Magnetic resonance imaging (MRI), photograph during surgery, macroscopic and microscopic features of the abscess. (A) On T1-weighted contrast-enhanced MRI, adhesions to the sigmoid colon and rectum are suspected. (B) Dense adhesions exist around the posterior wall of the uterus and sigmoid colon. (C) Photograph of the excised uterus. The endometrium is smooth with no inflammatory changes, and the abscess is located in the posterior wall of the uterus. (D) Microscopic feature (H.E.): marked infiltration of foamy histiocytes with clear lipid-containing cytoplasm, together with abundant lymphocytes and plasma cells, was observed.

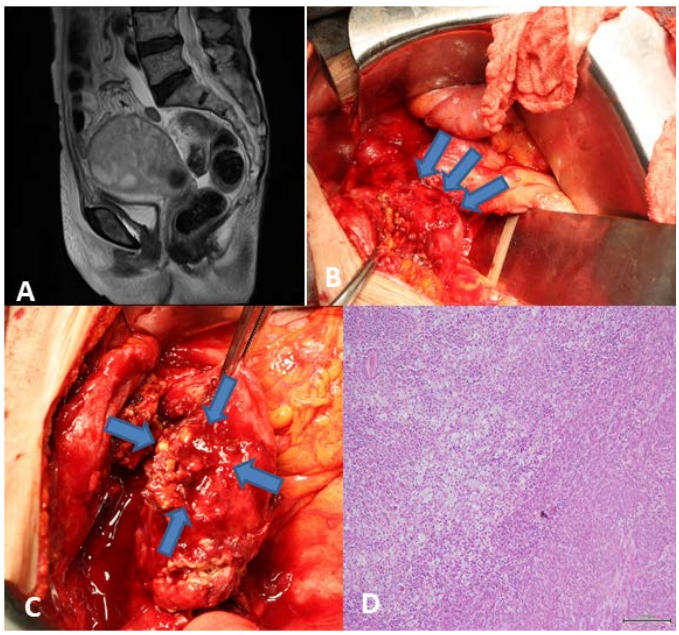

Figure 2. Case 2 Magnetic resonance imaging (MRI), photographs during surgery and microscopic feature of the abscess. (A) T2-weighted MRI revealed an irregularly shaped mass, 8 cm in diameter, which was localized in the posterior wall of the uterus. (B) Dense adhesions exist around the posterior wall of the uterus and intestine. (C) Uterine wall defect is observed on the uterine fundus. (D) Microscopic features (low power): The inflammation infiltrates deep into the myometrium (left) but do not reach the endometrium (right).

and 19.5 mg/dl, respectively. These findings were highly suggestive of abscess formation in the uterus. Total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed. A thick adhesion affecting the uterine fundus and intestine was found (**Figure 2B**). Adhesion lysis was performed, and uterine wall defect was observed on the uterine fundus (**Figure 2C**). The patient recovered without incident. Klebsiella pneumoniae was detected in culture from endometrial cavity. The pathological report of the uterus revealed xanthogranulomatous inflammation in the posterior myometrium and perimetrium, while there were no xanthoma cells except chronic inflammation in the endometrium and bilateral adnexae (**Figure 2D**).

3. Discussion

Xanthogranulomatous inflammation of the female reproductive organs is uncommon, and most reported cases are basically limited to the endometrium, ovaries, and fallopian tubes (tubal abscess) [3]-[16]. Only a few cases of xanthogranulomatous inflammation involving the myometrium and peritubular area have been reported. The present case is interesting because the affected myometrium was replaced by lipid-laden foamy macrophages and polymorphonuclear leukocytes, and xanthogranulomatous inflammation was observed in and around the myometrium, which further extended into the pelvis, resulting in uterine perforation. To the best of our knowledge, only two cases of xanthogranulomatous inflammation of the myometrium without endometritis have been reported; Liao *et al.* reported a case in which xanthogranulomatous inflammation was histologically found only in the myometrium and extended tansmurally into the peritoneal cavity, while the endometrium and fallopian tubes simply showed acute and chronic inflammation [17]. Inoue *et al.* also reported a case of xanthogranulomatous inflammation with periuterine to myometrial invasion without endometritis or oviductitis. They suggested that one of the origins of xanthogranulomatous inflammation in the female reproductive organs is not only in the endometrium and adnexa but also in the peritoneum [18].

One of the main differential diagnoses of xanthogranulomatous inflammation is malignancy [9]. Indeed, endometrial hyperplasia and endometrial carcinoma have been detected in patients with xanthogranulomatous inflammation associated with endometritis [19]. Therefore, cytological and histological diagnosis is essential to rule out endometrial malignancy in cases of xanthogranulomatous inflammation.

The etiology of xanthogranulomatous inflammation involving the female reproductive organs is not yet fully understood. Proposed causes are obstruction of the cervix, intrauterine devices, intrauterine manipulation, and infection [19]. Multiple factors are possible. The recognized theory is the infection theory, which is supported by a clinical history of infection and bacteria such as E. coli, Proteus vulgaris, Bacteroides fragilis, and Salmonella typhi cultured and grown from affected tissues [16]. Although the presence of large numbers of lipid-laden

foam cells cannot be explained by the infection theory alone, some authors have argued that it is a chronic inflammatory process leading to tissue necrosis and the continuous release of lipids such as cholesterol from dead cells phagocytosed by macrophages, leading to xanthomatosis [16].

The cases considered herein developed lower abdominal pain and fever after endometrial cytology. It cannot be determined that endometrial cytology was associated with abscess formation. The microscopic spread pattern of xanthogranulomatous inflammation in these cases suggests that the origin is more likely to be the myometrium rather than the endometrium, fallopian tubes. Considering that *Klebsiella pneumonie* was detected in the uterine cavity and mild local inflammation of the endometrium and fallopian tubes was confirmed, Klebsiella pneumonie present in the uterine cavity may have caused the perimetrial abscess through the fallopian tubes or hematogenously because of damage to the endometrium by intrauterine manipulation.

4. Conclusion

If inflammatory lesions are found after intrauterine manipulation, the possibility of developing xanthogranulomatous inflammation should be considered, and if antibiotics are not effective, prompt surgical treatment can reduce morbidity and improve patient prognosis.

Authors' Contributions

Kyousuke Takeuchi prepared the manuscript and is responsible for the overall content as guarantor. Ai Yoshida, Makoto Sugimoto, Masayuki Fujita and Hiroki Morita reviewed the manuscript. All authors read and approved the final manuscript.

Ethics Approval and Consent to Participate

Written informed consent was obtained from the patient for the publication of this case report. Ethical approval is waived for the case report.

Data Availability

Data are available on request (mail to kyousuket@dolphin.ocn.ne.jp).

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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