



Case report

Mimicking uterine malignancy: Pelvic actinomycosis with giant uterine leiomyoma



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ABSTRACT

Pelvic actinomycosis is a rare disease which is hard to be distinguished from other diseases such as malignant tumors and tuberculosis due to its nonspecific clinical signs and symptoms. If pelvic actinomycosis can be diagnosed preoperatively, the patients can be cured with antimicrobial therapy avoiding surgery. It is especially of concern to distinguish pelvic actinomycosis from pelvic mass, if there is a history of intrauterine device use. We report a case of pelvic actinomycosis that was diagnosed after the postoperative pathology of a suspected uterine malignancy.

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Introduction

Actinomycosis is a rare, chronic, suppurative granulomatous disease caused by filamentous Gram-positive anaerobic bacteria actinomyces [1]. Actinomyces often reside as colonizer on the mucous membranes of human oral cavity, gastrointestinal tract and female reproductive tract. Usually, actinomyces cannot cross the mucosal barrier of the above body parts [2]. But tissue injuries caused by trauma, surgery, or foreign body, can break the mucosal barrier and then lead to clinical infection. The clinical manifestations of actinomycosis are chronic infection, suppurative inflammation, and formation of fistulae and sinuses [3].

About 20 % of reported cases of actinomycosis is abdomino-pelvic actinomycosis [4]. Pelvic actinomycosis most often associate with intrauterine device (IUD) use [5]. The clinical symptoms of pelvic actinomycosis include fatigue, fever, weight loss and lower abdominal pain, sometimes associated with a palpable mass. Therefore, it is often misdiagnosed because it can mimic other conditions such as malignant tumor and tuberculosis [6]. It is estimated that less than 10 % of patients are diagnosed preoperatively, most of the cases were confirmed by exploratory laparotomy for suspected malignancy [1]. If pelvic actinomycosis can be

diagnosed preoperatively, the patients may be cured with antibiotic therapy, and can be avoided extensive extirpative surgery, potentially preserving fertility [7].

Although rare, a high level of clinical suspicion is needed to diagnose pelvic actinomycosis early in patients with chronic inflammatory disease and pelvic mass [1]. Preoperative diagnosis of pelvic actinomycosis is difficult, but clinicians should be aware of its unusual presentations and its ability to mimic malignancy. Several case reports had been previously presented pelvic actinomycosis to mimicking pelvic malignancy [7,8], however, there are few reports of pelvic actinomycosis penetrating the abdominal wall. Here, we report a case of pelvic actinomycosis with abdominal wall ulcer that was diagnosed after the postoperative pathology of a suspected uterine malignancy.

Case report

A 48-year-old woman presented to the oncological surgery outpatient department with to lower abdominal pain and an abdominal wall ulcer. She had a history of weight loss of 5 kg during the preceding 6 months with no history of diabetes mellitus or hypertension and had used an intrauterine device (IUD) for 20 years. She was afebrile and no vaginal bleeding. Physical examination revealed that the lower abdomen was swollen, and the two ulcers could be seen in the left lower abdomen and under the umbilicus, respectively. The skin around the ulcers was erythematous (Fig. 1). Laboratory findings revealed hemoglobin with a decrease (8.4 g/dL), normal leukocyte (8230/ μ L) and there

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Fig. 1. Skin defects.

was a normal carcinoembryonic antigen. Transvaginal ultrasound showed that the deep ulcer around the uterus and abdominal wall was hypoechoic, with consideration of infection granuloma formation, uterine cavity heterogeneous strong echo did not exclude malignant lesions. Abdominal and pelvic computed tomography (CT) plain scan showed that the lower abdominal wall was thickened with a large in the lower abdomen and pelvis with indistinct margins, and no obvious abnormality was seen in the liver and pancreas. Besides, there was without ascites (Fig. 2). Abdominal pelvic cavity magnetic resonance imaging (MRI) showed an intrauterine mass, considering the possibility of malignancy measuring 14.7 cm × 9.5 cm × 6.2 cm, but not excluding granulomatous process.

The biopsy of the ulcer in the lower abdomen showed inflammatory granulation tissue. An ultrasound guided puncture biopsy of uterine and pelvic masses suggested a myofibroblast tumor or smooth muscle-derived tumor. Abdominal and pelvic mass excision, abdominal wall granuloma excision, hysterectomy and salpingo-oophorectomy were performed. Postoperative paraffin pathology suggested that the abdominal and pelvic masses were actinomycosis with surrounding tissue fibrosis, uterine leiomyoma with hemorrhage, and chronic tubal inflammation (Fig. 3).

The patient received a diagnosis of pelvic actinomycosis infiltrating abdominal wall with uterine leiomyoma. The patient received piperacillin/tazobactam combined with metronidazole intravenously. The incision of patient healed by 20 days after operation (Fig. 4). She was discharged 5 months after the operation and computed tomography of the pelvic cavity was reexamined and no recurrence was found.

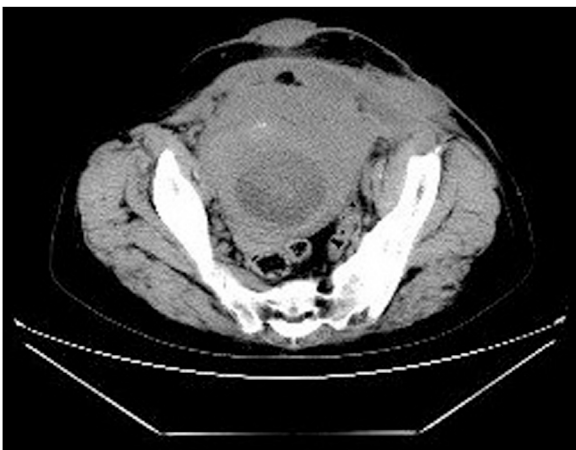


Fig. 2. Computed tomography of the pelvis showing large pelvic soft tissue mass invading the surrounding peritoneum.

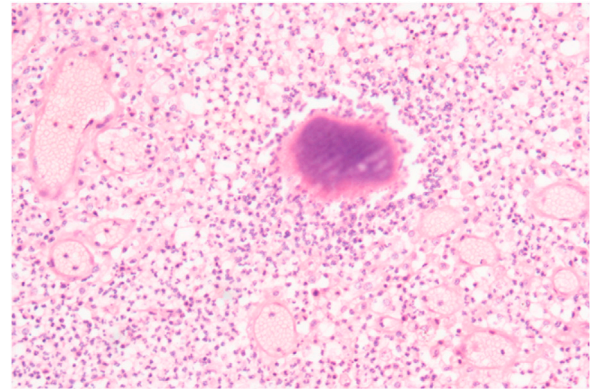


Fig. 3. Multiple lesions containing sulfur granules characteristic of actinomycosis.



Fig. 4. Postoperative incision healing.

Discussion

Actinomycosis is a rare and chronic granulomatous disease caused by filamentous Gram-positive anaerobic bacteria. The incidence rate of Actinomyces-like organisms in cervical smears was 0.26 % at a single university hospital [9]. A pelvic actinomycosis is associated with IUD uses, and about 7% of IUD users can be found to harbor Actinomyces-like organisms on staining. However, the pap smear diagnosis of actinomycosis lacks specificity, and only half patients are culture positive [10]. Pelvic actinomycosis may have no specific clinical symptoms such as fever, weight loss and lower abdominal pain and is often misdiagnosed because it can mimic malignancy and tuberculosis. Nevertheless, pelvic actinomycosis is actually treatable and curable if patients are appropriately managed, and some patients can avoid surgery.

In this case report, the patient was treated for abdominal pain accompanied by abdominal wall ulcer. CT examination showed that there was a large tumor in the pelvis, there were no ascites or lymph node involvement. We did not exclude the possibility of malignant tumor in the pelvis under the guidance of ultrasound. The tumor was punctured and biopsied and no malignant tumor cells were found. Because it was difficult to diagnose the nature of the mass in the pelvis before the operation, we performed laparotomy to explore the tumor in the pelvis. After the operation, the pathology was confirmed as actinomycosis. After anti-inflammatory treatment, the patient was discharged from hospital.

Several case reports and review had been previously presented this disease to mimicking pelvic malignancy [7,11,12]. Most of the cases were diagnosed by exploratory laparotomy for suspected

malignancy, it has been estimated that less than 10 % of cases are diagnosed preoperative [1]. For example, Sofia et.al [7] reported a case of pelvic actinomycosis mimicking ovarian malignancy diagnosed postoperatively. The patient had no complaints, a pap smear was obtained and the results were normal, the MR showed a retrouterine mass measuring 6 cm × 5 cm × 5 cm, which involved the distal left ureter, resulting in left hydronephrosis. The surgical resection was performed, and final histopathological revealed left tubo-ovarian actinomycosis with active chronic inflammation and abscess formation. Both this case and our case have a history of IUD use, and the mass is highly suspicious of pelvic malignancy. Morland et al. [8] reported that a 53 year old woman with a 3-month history of left inguino-crural and lumbar pain with weight loss. There was a history of IUD. Physical examination revealed a left adnexal mass measuring diameter 13 cm. Laparoscopy bilateral salpingo-oophorectomy and psoas biopsy were performed, and the diagnosis of pelvic actinomycosis was confirmed by histopathology. The third-generation cephalosporin was given 6 weeks after operation. There are a few significant differences between our case and previous cases. First, there were two ulcer foci on the skin of the abdominal wall, the texture is crisp, exudate is visible, similar to granulation tissue, but the patient has no fever. Second, the imaging examination showed that the skin ulcer foci were closely related to the pelvic masses. MRI considered the possibility of malignant tumors from the pelvic. Third, most of the cases can be cured by high doses of penicillin for 6–12 months. We used piperacillin-tazobactam and metronidazole for two weeks and then oral moxifloxacin was given for one month. No recurrence was found 5 months after surgery.

Author statement

Actinomycosis is a rare, the locally infiltrative pelvic mass is clear with the boundary of surrounding tissue, and the possibility of pelvic actinomycosis is highly suspected in patients with a history of IUD. The antibiotic treatment and the performance of surgery is crucial to cure the disease.

Availability of data and materials

The datasets used and analysed during the current study are available from the corresponding author on reasonable request

Authors' contributions

Xudong Song, Chuntao Wu, Ruiwei Li and Hongcheng Zhu treated the patient, Changzai Li and Pan Zhang wrote the report. Chunmei Ma, Jie Lv and Guimei Jiao review and edited. Jinji Zhang: supervision & review.

Ethics approval and consent to participate

This study was approved by the Ethics Committee of North China University of Science and Technology Affiliated Hospital (Tangshan, China), and was conducted according to the guidelines put forth in the Declaration of Helsinki.

Patient consent for publication

The patient provided written informed consent for the publication of any associated data.

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Declaration Competing Interest

The authors declare that they have no conflict of interests.

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