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A Case Report of Pelvic Actinomycosis and a Literature Review

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Conflict of interest: None declared

Patient: Female, 54-year-old
Final Diagnosis: Actinomycete
Symptoms: Tenderness
Medication: —
Clinical Procedure: Antibiotics • surgery removal
Specialty: Infectious Diseases

Objective: Rare disease

Background: The aim of this study was to report the clinical diagnosis and treatment of a case of pelvic actinomycosis in our hospital and provide a review of recent literature.

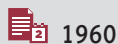
Case Report: The patient was a 54-year-old woman who was admitted to our hospital due to “bilateral lower abdominal tenderness accompanied with anorexia and vomiting for 3 months”. After admission, a variety of imaging examinations found pelvic space-occupying lesions, which were considered as malignant. She underwent surgery and pelvic actinomycosis was diagnosed by postoperative pathology. Postoperatively, she was treated with a high-dose sufficient course of penicillin (20 million U, iv gtt) for 14 days and she is currently under close follow-up for 1 year, with no recurrent symptoms.

Conclusions: Pelvic actinomycosis is rare and often forms mass invasion into the tissue structure around the pelvic cavity, which is easily misdiagnosed as ovarian malignant tumor. The criterion standard for diagnosing an infection is culture, with histopathology aiding the diagnosis.

MeSH Keywords: Decalcification, Pathologic • Genital Diseases, Female • Gynecology

Abbreviations: MRI – magnetic resonance imaging; CT – computed tomography; IUD – intrauterine device

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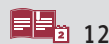
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Background

Actinomycetes are a group of gram-positive anaerobes or microaerobes, and the most common type of human pathogenic bacteria is *Actinomyces israelii* [1]. Actinomycosis is a rare, chronic, infectious disease characterized by infiltrative, suppurative, or granulomatous inflammation, sinus fistula formation, and extensive fibrosis [2]. It is relatively rare in clinical practice, with an annual incidence of 1/300 000 and a disease death rate of 0–28% [3]. Actinomycosis can be divided into facial type (50%), thoracic type (15%), abdominal and pelvic type (20%), and other types (15%). Because pelvic actinomycosis is rare and the symptoms are not typical, it often forms a mass invading the surrounding tissue structure, which is often misdiagnosed as a malignant tumor [4]. Here, we report a case of pelvic actinomycosis associated with use of an intrauterine device and diagnosed as ovarian malignant cancer by the imaging examination before surgery and by pathology. The clinical data were retrospectively analyzed and the relevant literature was reviewed to improve the understanding of actinomycosis among gynecologists.

Case Report

The patient was a 54-year-old woman. She was admitted to our hospital for bilateral lower abdominal tenderness accompanied by anorexia and vomiting for 3 months, and pelvic masses found 1 month before admission. Three months ago, without obvious inducement, bilateral lower abdominal tenderness

appeared, accompanied by loss of appetite, vomiting, constipation, no abdominal distension, and no vaginal bleeding. The ultrasonography examination showed a solid pelvic mass with an irregular shape, which occupied nearly all of the pelvic cavity. On physical examination, an irregular pelvic mass was felt, with no clear boundary and with positive tenderness. The pelvic magnetic resonance imaging (MRI) scan with contrast media (Figure 1) and the abdominal computed tomography (CT) scan (Figure 2) both confirmed these findings. We found that the patient had severe bilateral pyelectasis caused by the pelvic mass. After onset, she felt depressed and tired, with lack of appetite, poor sleep, and weight loss of 15 kg. A history of intrauterine device (IUD) implantation (removed 6 months ago) was reported. Preoperative laboratory test results of ovarian tumor markers were normal. The routine blood tests showed an increase of white blood cells ($13.28 \times 10^9/L$) and neutrophils (85.9%), a decrease of hemoglobin (70.00 g/L), and the C-reactive protein value was normal. Abnormal renal function was diagnosed, with urea nitrogen 11.68 (mmol/L) and creatinine 217.4 ($\mu\text{mol/L}$). Unfortunately, due to lack of fever, a blood culture was not performed. The colonoscopy and gastroscopy results were negative. Surgery was scheduled for tumor resection and tissue procurement for pathologic diagnosis. Before the surgery, she received ureteral stenting, and the creatinine level progressively decreased to normal.

During the laparotomy, no ascites and no obvious abnormalities were found in the peritoneum or the surface of the intestine of the middle abdomen, and there was a tight adhesion between the uterine and bilateral adnexa, ascending colon, and

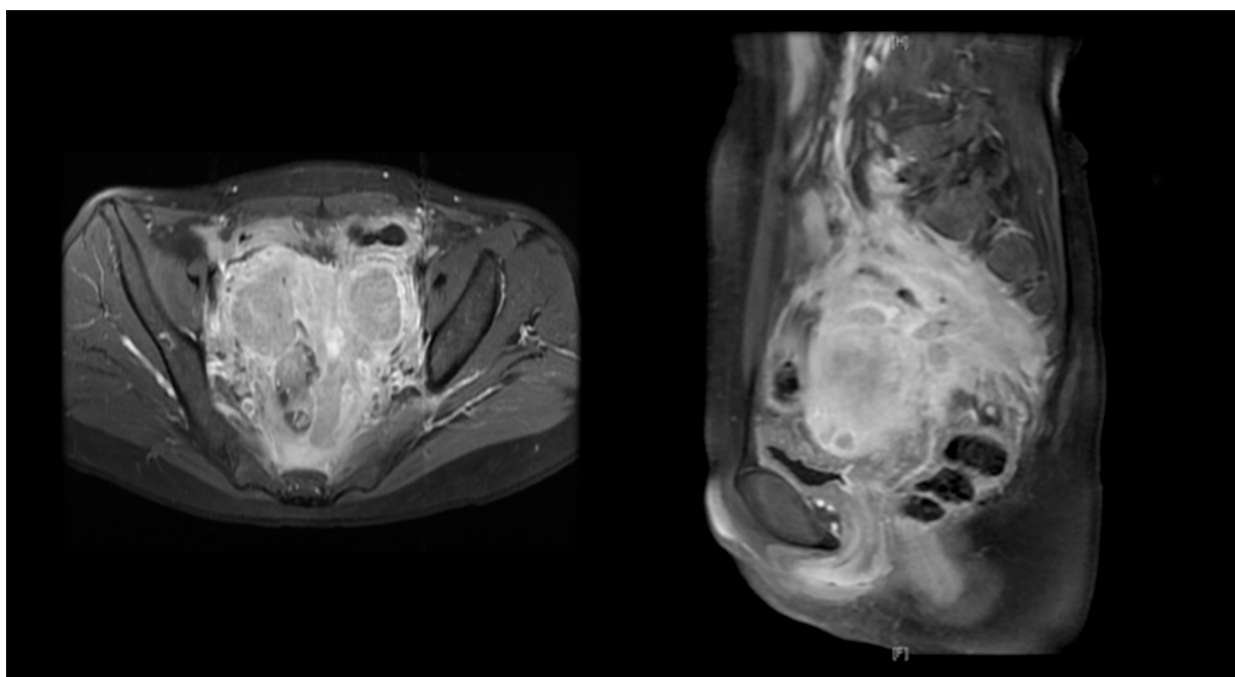


Figure 1. MRI with contrast media showed the pelvic mass, which was considered as malignant before surgery.

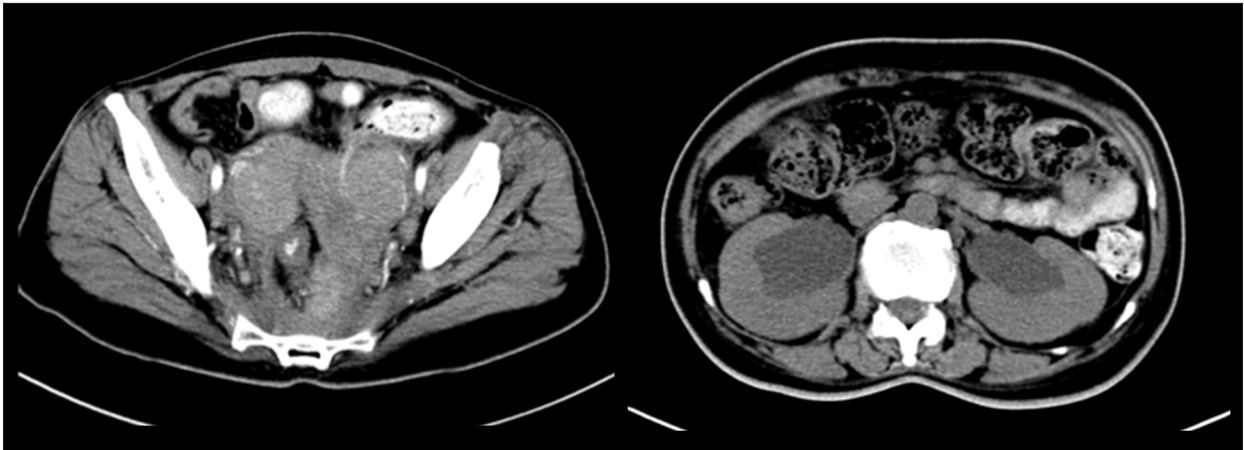


Figure 2. CT showing the pelvic mass and retro-renal pyelectasia.

sigmoid colon. During the surgery, we first separated the pelvic adhesion. We saw that the uterus was slightly enlarged and no signs of perforation were observed. Both ovaries were about 4 cm in diameter and solid; they were adhered and wrapped with double fallopian tubes. Both fallopian tubes were thickened and shortened by about 5–6 cm, with edema and stiffness. Pelvic organ adhesions usually show some necrotic organization, with no pus, no malodorous smell, and no typical caseous material. The pelvic peritoneal was thickened. After removal of the ovaries, the section view was full of yellow-gray nodules about 1–2 mm in diameter and there were similar tubal nodules, which were considered to be a pelvic infection. The frozen pathology during surgery confirmed the diagnosis was benign. Therefore, we decided to perform total hysterectomy with bilateral adnexectomy, and a pelvic drainage tube was placed to remove fluid after the surgery. Pathologic examination showed that the histologic features of the tumor and its immunohistochemical profile supported the diagnosis of pelvic actinomycosis (Figure 3), and results of gram stain was gram-positive rods. Postoperatively, she was treated with a high-dose sufficient course of penicillin (20 million U, iv gtt) for 14 days and she is currently under close follow-up for 1 year with no symptoms.

Discussion

Pathogenesis

Actinomycetes do not cause disease in normal parasitic sites such as the oral cavity, nasopharynx, gastrointestinal tract, and female reproductive system. When there is immune system abnormality or tissue trauma, the tubular mucosa ruptures or the whole layer of the tubular cavity is broken, and the actinomycetes transfer to the submucosa and body cavity, causing disease under the synergistic effect of other bacteria [5]. Abdominal actinomycosis usually originates after diaphragm perforation,

which is more common in males than females [6]. In recent years, the incidence of female pelvic actinomycosis has been rising, which is mainly related to the increased placement of IUDs [7]. Even after the removal of an IUD, some patients may be affected because of various degrees of damage to the endometrium caused by stimulation from the IUD, intrauterine flora disorders, and insufficient blood supply to the endometrium [8]. Cobellis [9] stated that postmenopausal actinomycosis was more likely to occur, perhaps because endometrium infected by actinomycosis cannot be cleared of menstrual blood after menopause. All the reported cases of actinomycosis were individual cases. Galata [10] reported a case of simultaneously invasive abdominal wall and celiac actinomycosis accompanied by pelvic internal urethral obstruction in a patient with a history of IUD implantation, which was easily overlooked because the IUD had been removed 8 years before.

Clinical features

Pelvic actinomycosis can cause abscess, which can involve the bladder, rectum, sigmoid colon, and other pelvic organs [11]. The 3 most common symptoms of pelvic actinomycosis are abdominal pain, weight loss, and abnormal vaginal discharge. It is reported that 60% of patients also have fever, and the clinical manifestations are less severe than the lesion [11,12]. In severe cases, a sinus tract or fistula is formed, and solid masses with fixed and unclear boundaries are formed, and even “frozen pelvis” can occur. Patients with elevated C-reactive protein, decreased hemoglobin, increased erythrocyte sedimentation rate, and slightly increased CA125 can have renal pelvis dilation or hydronephrosis. It has been reported that pelvic actinomycosis most commonly involves the diaphragm, cecum, colon, stomach, liver, pancreas, rectum, pelvic cavity, and abdominal wall [5]. In our patient, typical pelvic and abdominal adhesion lesions were observed during intraoperative exploration, and the intraoperative pathological examination mostly indicated abscessed wall tissues and inflammation [9].

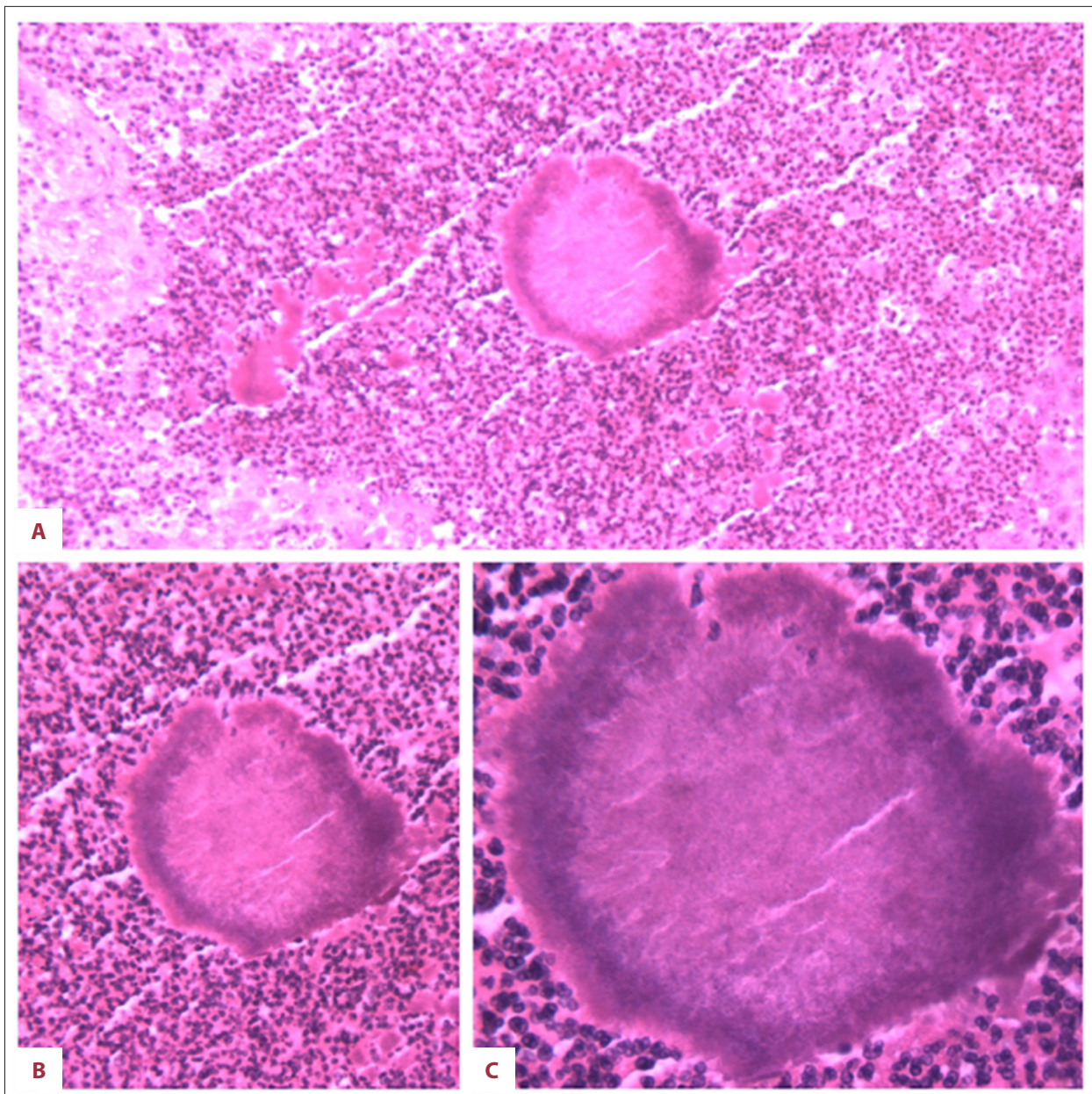


Figure 3. Actinomycete colony can be seen under 100× (A), 200× (B), and 400× (C) under an optical microscope, with surrounding neutrophil infiltration.

The clinical manifestations of our case were lower abdominal pain, anorexia, vomiting, abdominal mass, weight loss, anemia, and emaciation. The patient had a history of easily overlooked IUD implantation, which was removed half a year ago, and she had been menopausal for 4 years. The lesion involved the bilateral appendix and the pelvic wall, and was misdiagnosed as an ovarian malignant tumor before surgery.

Diagnosis points

According to the literature, the diagnostic principles of pelvic actinomycosis are: 1) Chronic suppurative inflammation with mass, extensive adhesion, and sinus and fistula formation; 2) Sulfur particles in the pus; 3) Extensive inflammatory infiltration, necrosis and abscess observed microscopically, with inflammatory granulation tissue hyperplasia, purple-red cloudy colony formation, and positive gram staining. CT and MRI imaging examinations lack diagnostic specificity, but can still be important in judging the involvement range of abdominal organs

and identifying the mass wall characteristics [5]. Postoperative pathological examination in our case showed a lesion with masses of radiating hyphae and granulomatous inflammation accompanied by foam cells.

Misdiagnosis

Pelvic actinomycosis has the clinical manifestations of invasive lesions, which are easily misdiagnosed as a malignant tumor and are difficult to diagnose preoperatively. It is reported that the rate of preoperative diagnosis is less than 10%, and most were diagnosed after the operation [5]. The reasons for our misdiagnosis in the present case were: 1) Lack of experience and knowledge of actinomycosis, and the patient mainly presented with space-occupying lesions in the abdomen and pelvis, and presented with cachexia; 2) Difficulty in differential diagnosis: this case did not have the typical clinical manifestations of infectious diseases, the symptoms and signs were non-specific, the preoperative tumor markers were normal, the positive rate of hematuria bacteria culture was low, and the clinical and imaging manifestations were similar to those of malignant tumors, leading to difficulty in differential diagnosis. CT and MRI imaging features of this disease are generally mixed pelvic air mass, invasion of surrounding organs, and lack of specificity, so it is difficult to distinguish between pelvic inflammatory diseases and malignant tumors.

Treatment principles

The preferred drug for treating actinomycosis is a high-dose sufficient course of penicillin, which is recommended to be given as an intravenous infusion of 18~24 million U per day for 2~6 weeks, followed by oral administration of 2~4 g/d for 6~12 months [5]. However, in recent years, individualized treatment has been emphasized, and the specific treatment plan depends on the lesion, the infected site, whether the patient is willing to undergo surgery, and the clinical and imaging response to treatment. A large dose and long course of treatment are needed to prevent recurrence, and the possibility of IUD implantation should be considered in gynecological diseases. Studies have shown that actinomycetes are sensitive to a variety of antibiotics, and sulfonamides, erythromycin, doxycycline, tetracycline, and other drugs can be used when there is penicillin allergy or antibiotic resistance [5-7]. In addition, it has been reported that levofloxacin also has a therapeutic effect on actinomycosis [9]. In patients with rapid progression of actinomycosis, mixed infections are common and should be treated with broad-spectrum antibiotics. Patients with complicated actinomycosis who have failed medical treatment and have abscess or sinus tract formation need surgical treatment. Although inflammation is highly suspected to

be the cause of the lesion during intraoperative exploration, the possibility of tumor co-infection should not be excluded. For safety reasons, the lesion should be extensively and thoroughly cleared during surgery [5,9,11]. At present, it is considered that patients with masses should undergo surgery as soon as possible, while avoiding unnecessary injury to the patients, so as to make a clear diagnosis. On the basis of correct preoperative and intraoperative diagnosis, surgery can narrow the range of lesions and increase the penetration of drugs, thus shortening the treatment process of drugs and reducing the dosage, in order to enhance the efficacy and reduce the recurrence rate. The necrotic tissue and fistula can be resected to relieve compression and obstruction. To avoid misdiagnosis of malignant tumors and over-treatment, it is advisable to take samples for examination. It should be noted that surgery should be thorough, as incomplete surgery or drainage is the root cause of postoperative recurrence. Fiorino [8] summarized 92 cases of pelvic actinomycosis, including 16 cases of intestinal resection and 2 cases of bladder resection. Using surgery combined with large-dose, long-course antibiotic treatment, the cure rate is about 90%.

Our patient was diagnosed as having a malignant disease based on the clinical features and examination. Due to the seriousness of her state of illness, we performed surgery, with ureteral stenting preoperatively. During the operation, we found an inflammatory disease with a benign result of rapid-freezing pathological examination. Postoperative pathology identified pelvic actinomycosis. Our experience with this patient shows that pelvic actinomycosis very difficult to distinguished from a malignant tumor, and the history of IUD use should be considered before treatment.

Conclusions

Pelvic actinomycosis is a clinically rare, insidious, purulent, infectious disease. Due to atypical symptoms, it often forms a mass invasion into the tissue structure around the pelvic cavity, which is easily misdiagnosed as a malignant ovarian tumor. Therefore, in patients with a history of IUD implantation and pelvic mass, actinomycosis should be excluded when a malignant pelvic tumor is suspected. Rapid-freezing pathological examination of multiple intraoperative samples can definitively diagnose the disease. The criterion standard for diagnosing an infection is culture, with histopathology aiding the diagnosis.

Conflict of interests

None.

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