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Abdominopelvic actinomycosis in three different locations with invasion of the abdominal wall and ureteric obstruction: An uncommon presentation



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ABSTRACT

INTRODUCTION: Actinomycosis is a rare chronic infectious disease caused by Gram-positive anaerobic bacteria that normally colonize the bronchial system and gastrointestinal tract in humans. The most common diseases associated with actinomycosis are orocervicofacial, thoracic and abdominal infections involving *Actinomyces israelii*. Due to its rarity, its various clinical presentations and often-infiltrative characteristics in radiological imaging, it can easily be mistaken for other clinical conditions, including malignancy.

PRESENTATION OF CASE: We present an uncommon case of extended abdominopelvic actinomycosis with infiltrative lesions in multiple locations, including an abscess in the abdominal wall and ureteric obstruction, which underwent successful surgical and subsequent long-term antibiotic therapy.

DISCUSSION: To our knowledge, such a combination of different sites of manifestation has not yet been reported for actinomycosis in the presence of an IUD. Possible differential diagnoses included diverticulitis with covered perforation, pelvic inflammatory disease, tuberculosis and inflammatory bowel disease. The possibility of a malignant process required radical resection. As in most cases of actinomycosis, diagnosis could not be established with certainty until postoperative pathology investigation.

CONCLUSION: A rare actinomycosis infection should be considered in patients with a non-specific pelvic mass and atypical abdominal presentations, especially if a previous history of IUD usage is known.

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1. Introduction

Actinomycosis is an uncommon chronic infectious disease largely unknown to most clinicians. It is caused by a group of anaerobic, microaerophilic Gram-positive bacteria that normally colonize the human bronchial system and gastrointestinal tract. Infection often causes infiltrative inflammation, florid abscess formation, chronic granulomatous lesions, fistulation and tissue fibrosis [1]. Clinical presentations in the orocervicofacial area, the thorax and the abdominal cavity are most frequently reported. The most common type causing disease in humans is *Actinomyces israelii* [2]. Due to its rarity, the unspecific symptoms and the tendency to appear as a mass invading surrounding structures,

actinomycosis is often mistaken as a malignant tumor [3]. We present a case of extended actinomycosis in three different abdominopelvic locations induced by a neglected intrauterine contraceptive device (IUD) that was successfully treated by surgical intervention and long-term antibiotic therapy.

2. Presentation of case

A 54-year-old woman presented in a reduced general state of health at the emergency department of our hospital with diffuse abdominal pain and dyspnea for several hours. She reported constipation and deterioration of her general condition for 1 week. Initial examination showed a tachyarrhythmia up to 175/min and a body temperature of 40 °C. As pre-existing conditions, atrial fibrillation and lung embolism after a Caesarean section 20 years ago were known. The patient was not on any medication. Weight loss was not a symptom. Routine laboratory tests showed elevated D-dimer levels (> 32 mg/l), signs of systemic inflammation (CRP

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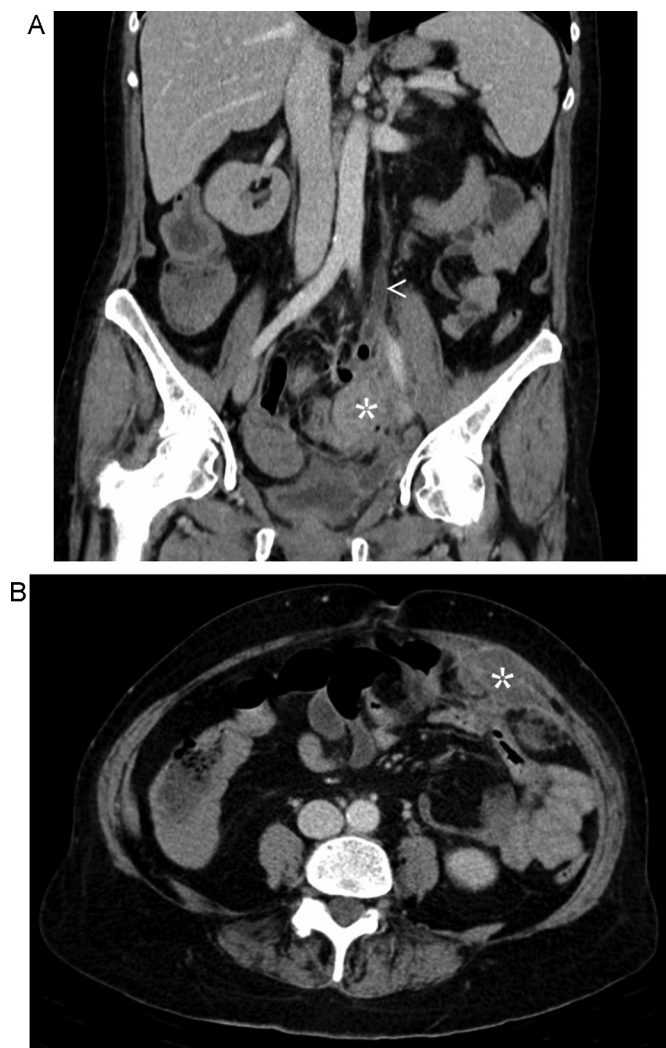


Fig. 1. (A) Computed tomography image obtained from the mass (*) in the lower abdomen and pelvis. Note the invasion of nearby anatomic structures and the dilated left ureter (>). (B) Computed tomography image demonstrating the abscess (*) in the left upper abdomen.

280 mg/dl), anemia (plasma hemoglobin 9.7 mg/dl), electrolyte disorder (plasma potassium 3.0 mmol/l) and impaired liver and renal function (INR 1.43, plasma creatinine 1.14 mg/dl). Urinalysis revealed elevated leucocyte and erythrocyte levels.

For further investigation, computed tomography (CT) scans of the thorax and the abdomen were obtained. Imaging of the thorax showed no signs of peripheral or central lung embolism. The CT scan of the abdomen revealed a pelvic mass with trapped air that was encroaching on the uterus, the adnexa and the sigma (Fig. 1A). The mass compressed the left ureter with consecutive urinary obstruction. There were no signs of an iliac vein thrombosis. In addition, a lesion indicative of an abscess formation was apparent in the left upper abdominal wall (Fig. 1B). Directly adjacent was an intraabdominal abscess involving the small bowel and transverse colon.

Ureteral stenting of the left ureter was performed. Gynecological examination of the patient revealed no pathological findings except a mild colpitis. However, an intrauterine contraceptive device that had not been checked for the last eight years was removed and empirical therapy with intravenous antibiotics was initiated.

Ultrasound controlled drainage of the lesion in the left abdominal wall was performed and revealed purulent discharge. Sigmoidoscopy showed no signs of diverticulitis or perforation but

proved a complete stenosis of the sigma, allegedly by external compression. While no sign of recovery occurred and laboratory testing indicated septic illness, the indication for urgent exploratory laparotomy was confirmed, and informed consent was obtained from the patient. Intraoperative findings confirmed the radiographic diagnosis of a conglomerate tumor in the left lower abdomen. Ureterolysis of the left ureter, left ovariectomy and extended radical left hemicolectomy with resection of the transverse colon was performed. The abscess in the left upper abdominal wall was connected to the transverse colon; however, drainage produced only pus. The small bowel that was adjacent to the formation near the transverse colon was released, and all abscesses including the lesion in the left abdominal wall were lanced. The rectal stump was closed, and a colostomy of the right flexure was created. In the postoperative course, the patient initially became better quickly. On the 5th post-operative day, symptoms became aggravated again and radiographic imaging of the chest showed free air. Relaparotomy was performed, which revealed a fresh peritonitis due to a dot-like lesion in the small intestine. This perforation was in the area that was released from the colon. Segmental small bowel resection (10 cm) with end-to-end anastomosis was necessary. The further postoperative course was uneventful.

Meanwhile, pathological investigation of the resection specimen demonstrated abdominopelvic actinomycosis (Fig. 2). Subsequent antibiotic therapy consisted of intravenous extended spectrum β -lactam antibiotics for 3 weeks and oral maintenance therapy for 3 months. The patient was discharged in good medical condition and was followed up on at our gastroenterology outpatient clinic.

3. Discussion

Actinomycosis is a rare disease. The most common form is orocervicofacial actinomycosis, which accounts for approximately 50% of reported cases and usually follows dental trauma or manipulation [4]. Manifestations in the thoracic or abdominal cavity are also common. Abdominal actinomycosis traditionally originates from perforated appendicitis [5] and is more often found in men than women [1]. In the recent past, abdominal actinomycosis has taken on greater significance in females because an increasing number of cases associated with the use of intrauterine contraceptive devices (IUD) have been reported [3]. Actinomyces do not populate the female genital tract in the absence of an IUD, and a preceding mechanical breakdown of the mucosal barrier is required for infection [1].

We present an extraordinary case of disseminated abdominopelvic actinomycosis with infiltration of the abdominal wall presumably caused by a neglected IUD. The involvement of multiple separate intraabdominal and pelvic lesions is very rare. In the literature, there is one report of a case where a patient with abdominal wall actinomycosis developed two secondary intraabdominal abscesses after surgical evacuation of the initial lesion [6]. Likewise, there is little literature on the subject of invasion of the abdominal wall in cases of actinomycosis originating from the prolonged use of an IUD [6,7]. Secondary ureteric obstruction has been described before in pelvic actinomycosis and is a known complicating factor [8]. To our knowledge, the combination of these three separate sites of manifestation in a single case—abdomen, pelvis with ureteric obstruction and the abdominal wall—has not been reported yet for actinomycosis in the presence of an IUD. It is typical that the aggressive appearance of the disease, as in our patient, makes correct preoperative clinical diagnosis nearly impossible. Differential diagnoses included diverticulitis with covered perforation, pelvic inflammatory disease, tuberculosis and inflammatory bowel disease. Because of the

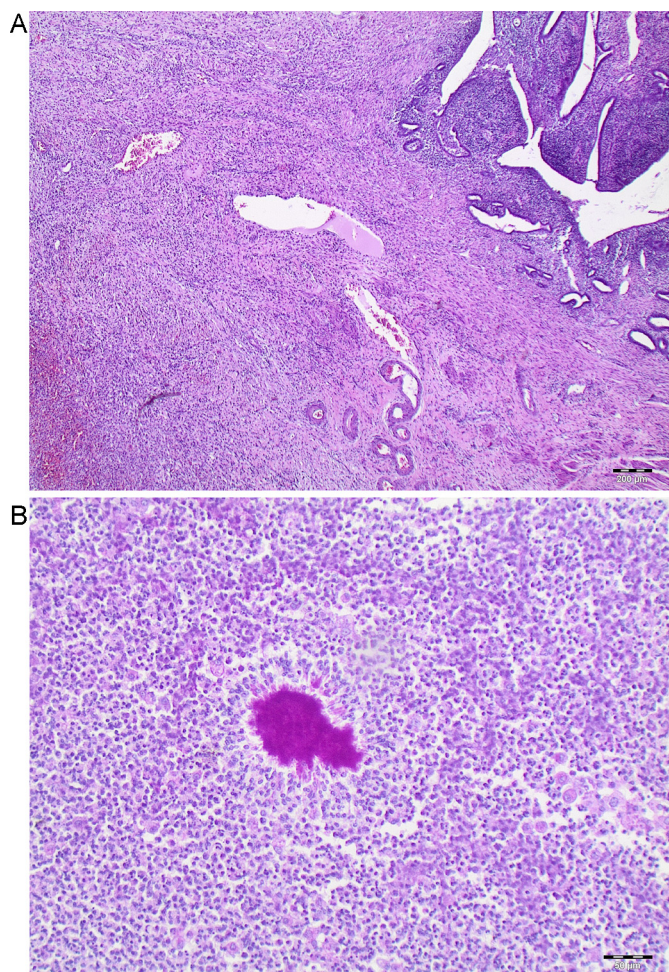


Fig. 2. (A) Histology image of the retrieved material showing the fallopian tube with inflammatory changes of the wall (A); 20 \times ; HE. (B) Characteristic histological presentation of actinomycosis with actinomycotic granules composed of radiating filaments with a dense granular core, surrounded by inflammatory cells composed of a mixture of neutrophils and eosinophils (B); 100 \times ; PAS.

infiltrative appearance in radiological imaging, the possibility of a malignant process had to be taken into account. The indication for urgent exploratory surgery was confirmed based on the clinical condition of the patient. Even in a patient displaying no signs of septic illness, elective surgery would have been recommended due to suspected malignancy. During surgery the situs seemed to be caused by inflammation; however, radical resection was performed for safety reasons. As in most cases of actinomycosis, a concrete diagnosis could not be established until postoperative pathology investigation.

Regardless of the operative treatment, subsequent high dose antibiotic therapy had to be carried out to ensure adequate drug penetration and to prevent delayed relapse [1]. In general, β -lactam antibiotics combined with β -lactam inhibitors are the treatment of choice. For penicillin-allergic individuals, erythromycin and tetracycline have been reported to be effective alternatives [9]. However, susceptibility can vary between actinomyces species [10]. The optimal duration of treatment remains unclear. Previous recommendations varied between 6 and 12 months, but short-term treatments for 2–6 weeks after surgery have been successful [4]. The length of treatment has to be individualized and should be based on the initial burden of disease, the completeness of surgical resection and the response to treatment.

4. Conclusion

Abdominopelvic actinomycosis is an uncommon and insidious suppurative infection that still poses a great diagnostic challenge. It leads to infiltrative pelvic and abdominal mass lesions that can mimic a variety of conditions from malignancy to acute inflammatory pathologies and may be complicated by the development of lesions on the abdominal wall or by ureteric obstruction. In the majority of cases, the diagnosis is only confirmed after surgery by pathology or microbiological examination of resection specimens. However, the possibility of a rare actinomyceal infection should be considered in patients with a non-specific pelvic mass and atypical abdominal presentations, especially if a previous history of IUD usage is known.

Conflicts of interest

No conflicts of interest.

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Ethical approval

None.

Consent

Written and signed consent has been obtained from the patient.

Author contribution

Galata C.L. was involved in treatment of the patient and wrote the manuscript.

Vogelmann R. was the gastroenterologist responsible for antibiotic treatment and follow-up and contributed to the manuscript.

Gaiser T. was the investigating pathologist and contributed to the manuscript (histology images).

Post S. critically read and corrected the manuscript and supervised treatment.

Horisberger K. performed the operation and contributed to the manuscript.

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