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Intraabdominal actinomycosis resulting in a difficult to diagnose intraperitoneal mass: A case report

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ABSTRACT

INTRODUCTION: Actinomycosis is a chronic suppurative granulomatous disease caused by *Actinomyces israelii*. Preoperative confirmed diagnosis is very difficult, so most cases are diagnosed preoperatively as malignant tumors. We report a case of intraabdominal actinomycosis which was difficult to diagnose preoperatively.

PRESENTATION OF THE CASE: A woman, 60 years old, experienced discomfort in her lower right abdomen. She complained of nausea and anorexia and visited our hospital. Laboratory blood tests, abdominal CT, and abdominal MRI led to a diagnosis of a uterine sarcoma or primary intestinal mass, and she underwent surgery. Her histopathological diagnosis was intraabdominal actinomycosis.

DISCUSSION: Actinomycosis is a chronic purulent granulomatous inflammation caused by *Actinomyces israelii*. No clinical symptoms or laboratory findings are characteristic of abdominal actinomycosis, so this disorder is very difficult to diagnose preoperatively. Therefore, many cases are diagnosed as malignant tumors and undergo surgery. After surgery, long-term antibiotic treatment (penicillin) is usually administered.

CONCLUSIONS: We reported a case of intraabdominal actinomycosis that resulted in a difficult to diagnose intraperitoneal mass. When a large intraperitoneal mass is found, actinomycosis needs to be included as one of differential diagnoses.

1. Introduction

Actinomycosis is a chronic suppurative granulomatous disease caused by *Actinomyces israelii*. It is reportedly caused by invasion from sites of damaged mucous membranes arising from factors such as appendicitis, diverticulitis, and uterine contraceptive devices. This disease is very difficult to diagnose preoperatively, and tends to be diagnosed as a malignant tumor and treated surgically. The correct diagnosis of a chronic suppurative granulomatous disease is made based on histopathological findings of the surgical specimen.

We report a case of intraabdominal actinomycosis that was difficult to diagnose preoperatively. This work has been reported in line with the SCARE criteria [1].

2. Presentation of case

A 60-year-old female patient experienced discomfort in her lower right abdomen beginning in August 2016 and her symptoms did not improve. By the end of November 2016, she was experiencing nausea and anorexia, and she visited our hospital.

On physical examination, a mildly tender and solid mass, sized 10 cm and with no mobility, was palpable in the lower right abdomen. No muscular defense or rebound pain were detected. Blood test findings revealed a white blood cell count of 8040/μl, hemoglobin of 11.9 g/dl and C-reactive protein (CRP) of 1.75 mg/dl. These findings indicated mild anemia and a mild inflammatory reaction. Other tests and tumor marker assessments gave results within the normal range.

Abdominal enhanced computed tomography (CT) revealed an extensive edematous wall thickening of the ileocecal area (Fig. 1). Abdominal magnetic resonance imaging (MRI) showed a large tumor with a long diameter of 9.6 cm located from the ileocecal area to the posterior cervical side of the uterus (Fig. 2). Based on these

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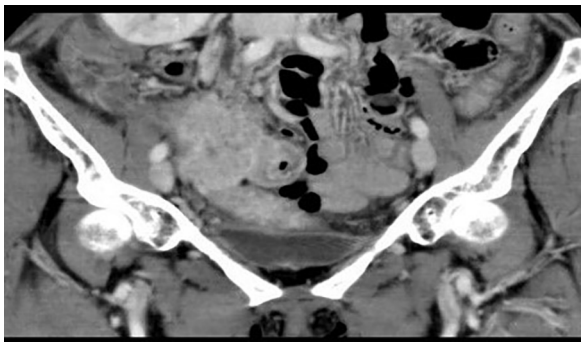

Fig. 1. CT scan showing an extensive mass and the surrounding tissue, accompanied by a contrast effect in the abdominal cavity.

findings, we diagnosed a uterine sarcoma or ileocecal malignant tumor and we performed a laparotomy.

The intraoperative finding was that the large tumor occupying the pelvic cavity appeared to be a tumor derived from the intestinal tract rather than from the uterus. The right ovary and uterus were involved with the tumor. The right external iliac artery and vein also had tumor involvement, so we could not resect the entire mass. We performed an ileocectomy and a complicated excision of the right ovary and part of the uterus, but a part of the tumor remained in the peritoneal cavity. An excised specimen revealed only wall thickening with edema in the intestinal mucosa, and no tumor was evident (Fig. 3a and b). The histopathological findings supported a chronic suppurative granulomatous disease caused by *Actinomyces israelii*. A bacterial mass was confirmed in the tissue, and a radial corona, consisting of a club-like structure and a collection of neutrophils, was observed around the bacterial mass (Fig. 4). No lymph node metastasis was detected.

The patient was discharged on the 16th postoperative day without postoperative complications. After surgery, she was prescribed antibiotics for six months. A follow-up observation after the antibiotic treatment revealed no recurrence for one year after her operation.

3. Discussion

Actinomycosis is a chronic purulent granulomatous inflammation caused by *Actinomyces israelii*. Actinomyces species are anaerobic gram-positive bacilli that exist in the oral, gastrointestinal, and upper respiratory tracts of a healthy person. The favored locations for actinomycosis are the head and neck, chest, and abdomen, with reported frequencies of about 60, 20, and 20%, respectively. The overall incidences of registered cases of actinomycosis are decreasing, but cases of abdominal and genital actinomycosis are increasing in frequency [2–5].

Abdominal actinomycosis can arise from appendicitis, diverticulitis, and fish bones [4–8]. The infection due to Actinomyces can derive from damaged sites and intrauterine devices (IUDs) [4].

The clinical symptoms are only fever and abdominal pain. No characteristic findings are reported in laboratory blood tests, and radiological findings are non-specific. This disease is often diagnosed as an irregular mass by abdominal ultrasonography and as an invasive inflammatory mass by abdominal CT [9]. Endoscopic findings show a smooth surface of the mucosa and no ulceration and no specific image shows the submucosal tumor. Even a biopsy shows only nonspecific chronic inflammation [10].

Abdominal actinomycosis is very difficult to diagnose preoperatively. Confirmation of the diagnosis includes any one of the following: (1) detection of a bacterial mass in pus, (2) confirmation of a bacterial mass by histology, or (3) detection of *Actinomyces* spp.in culture. However, even the culture of specimens from biopsies of infected fowl can reveal few bacteria. As a result, many cases are diagnosed preoperatively as malignant tumors.

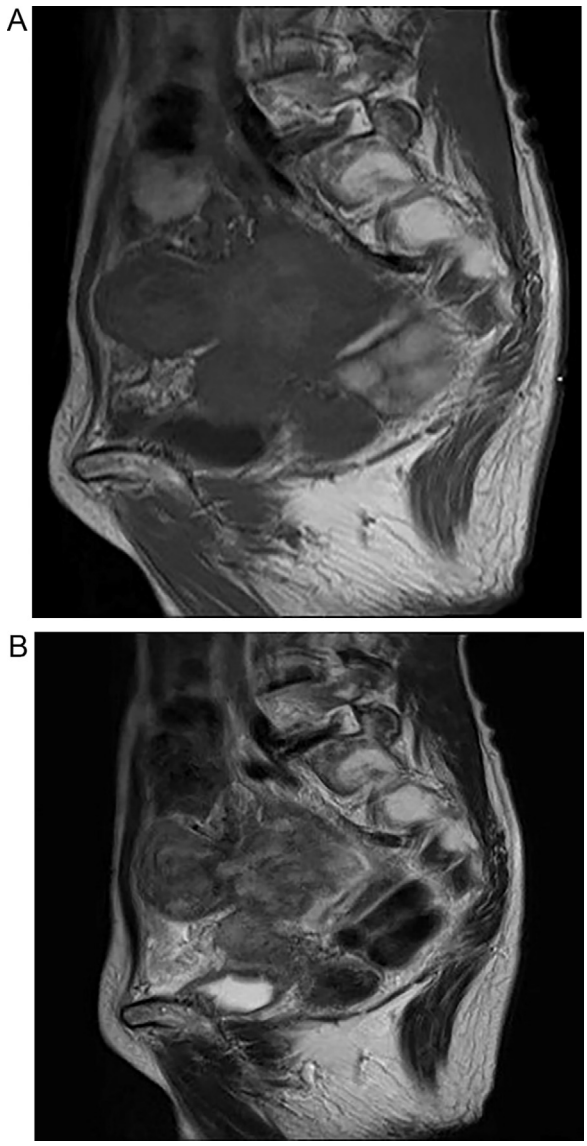

Fig. 2. The mass was seen with a low signal intensity on T1-weighted images (2A) and with an inhomogeneous signal intensity on T2-weighted images (2B). The boundary between the tumor and the uterus was unclear.

The treatment for abdominal actinomycosis is surgery and long-term antimicrobial administration, mainly penicillin [11–13]. Medication alone is not particularly effective because the lesion contains a lot of connective tissues and few blood vessels.

Surgical resection is usually performed because of the difficulty in confirming the diagnosis preoperatively and the suspicion of a malignant tumor [14]. This disease requires a long-term antibiotic administration because of the possibility of relapse after resection. Schtech reported that a duration of 1 year and 8 months of antibiotic administration was required [15], so long-term administration is necessary unless side effects emerge. However, no clear standards have been established for dosage or administration duration.

In our case, we performed surgical resection without endoscopic examination. However, endoscopic examination could be useful for a definitive diagnosis, because identification of the bacterial mass might be possible with a specimen from a biopsy. We also did not recognize any inflammation, such as appendicitis, diverticulitis, or a history of uterine contraceptive device insertion, etc. If a patient

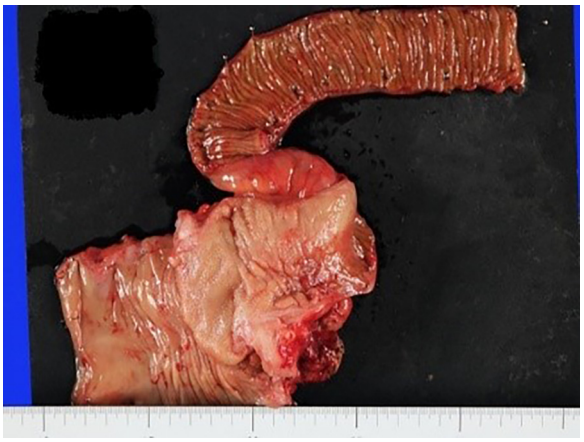

Fig. 3. Photographs showing the normal intestinal mucosa; no invasion into the ileocecal valve was observed. Cecal wall thickening showing a suspected cecal or appendiceal tumor.

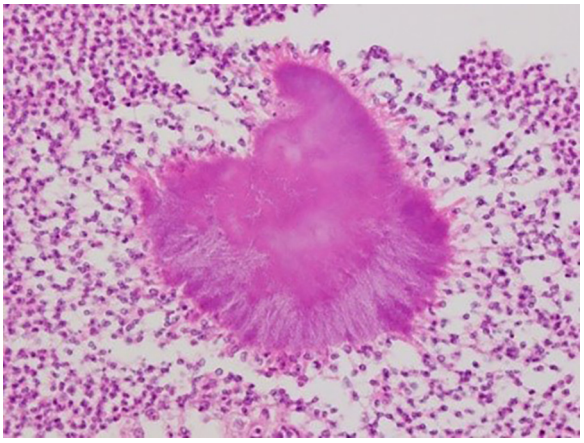

Fig. 4. The histopathological findings confirmed a bacterial mass in the tissue. A radial corona consisting of a crown structure and a collection of neutrophils was observed around the bacterial mass.

has a massive tumor with this history, abdominal actinomycosis should be considered.

4. Conclusion

We experienced a case of abdominal actinomycosis that was difficult to diagnose preoperatively. When a large intraperitoneal tumor is found, actinomycosis needs to be included as one of the differential diagnoses.

Conflict of interest

The authors declare no potential conflict of interest.

Funding

None.

Ethical approval

Ethical approval for a case report is not required by our institution.

Consent

Written informed consent was obtained from the patients for the information to be included in our manuscript. His information has been de-identified to the best of our ability to protect his privacy.

Author contribution

Each author participated in writing the manuscript and all agreed to accept equal responsibility for the accuracy of the content of the paper.

Guarantor

Naoto Tsujimura.

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