Case Report

Abdominopelvic Actinomycosis Mimicking Peritoneal Carcinomatosis: A Case Report

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Running title: Actinomycosis mimicking carcinomatosis
ABSTRACT

Actinomycosis is a rare chronic bacterial infection primarily caused by Actinomyces israelii. A 47-year-old woman presented to our clinic with a 1-week history of low abdominal pain. Preoperative imaging studies revealed multiple peritoneal and pelvic masses suggestive of malignancy. The primary tumor could not be identified despite further endoscopic and gynecological evaluation. On exploration for tissue confirmation, excisional biopsy of the multiple masses was performed because complete excision was not possible. Histopathological examination confirmed actinomycosis with multiple abscesses, and the patient was treated with antibiotics. We present a case of disseminated peritoneal actinomycosis mimicking malignant peritoneal carcinomatosis on imaging studies.

Key words: Actinomycosis, Carcinomatosis, Intrauterine device
INTRODUCTION

Actinomycosis is a rare chronic or subacute bacterial infection primarily caused by *Actinomyces israelii*, an anaerobic gram-positive or microaerophilic/capnophilic organism belonging to the *Actinomyces* genus [1]. Pre- and intraoperative diagnosis is challenging owing to the nonspecific clinical findings observed in patients, and the clinical presentation of actinomycosis can mimic that of malignancies [2]. We report a rare case of abdominopelvic actinomycosis mimicking peritoneal carcinomatosis of unknown primary origin.

CASE REPORT

A 47-year-old woman was referred to our clinic with a 1-week history of low abdominal pain and a palpable mass on abdominal examination. She denied a history of abdominal operations, but she reported intrauterine device (IUD) implantation 5 years prior to presentation. The patient’s vital signs were within the normal range. Abdominal examination revealed a hard mass in the left lower quadrant (LLQ) of the abdomen with no acute peritoneal signs. Blood tests were negative for leukocytosis or C-reactive protein (CRP) elevation, suggestive of systemic inflammation.

She was diagnosed with uterine myoma and a perirectal mass by pelvic magnetic resonance imaging performed at a local medical center. Gynecological examination at our hospital revealed no findings suggestive of gynecological malignancy; however, her serum cancer antigen (CA) 125 level was elevated (89 U/mL). The IUD was removed by gynecologist. Contrast-enhanced abdominopelvic computed tomography (CT) revealed a huge enhanced mass connected to the greater omentum in the left lower abdomen and a perirectal mass connected to the uterus, suggestive of peritoneal carcinomatosis (Fig. 1). Positron-emission tomography performed for diagnosis of the primary malignancy revealed a suspected carcinomatous lesion in the LLQ and multiple masses showing fluorodeoxyglucose uptake near the right lower quadrant and the perirectal area (Fig. 2). Endoscopic examination for assessment of alimentary tract malignancy did not reveal lesions suggestive of malignancy. Serum levels of carcinoembryonic antigen and CA19-9 were also within the normal range. Therefore, surgical exploration was performed for tissue confirmation.
Laparoscopic exploration was impossible because of a frozen abdomen secondary to the omental mass; therefore, median laparotomy was performed. Intraoperatively, we identified a firm mass, which was strongly adherent to the left lower parietal peritoneum. This mass was connected to the greater omentum around it. The mass projected from the posterior sheath of the rectus abdominis, and histopathological examination revealed it to be a focal nodular fibrotic lesion with central necrosis (Fig. 3A, 3B). A similar mass protruded from the right lower abdominal wall but did not involve the abdominal visceral peritoneum (Fig. 3C, 3D). The appendix in the vicinity of the mass appeared normal. Another firm mass was identified attached to the uterus on the pouch of Douglas; however, this mass did not invade the rectal wall (Fig. 3E, 3F). Frozen section biopsies from multiple sites revealed chronic inflammation with atypical cells; therefore, malignancy could not be excluded. The operation was abandoned because significant adhesions between the major vessels and intra-abdominal organs and the abdominal wall prevented complete resection.

The patient’s postoperative recovery was uneventful. Final histopathological findings included chronic active inflammation with multiple abscesses, dense fibrosis, multiple lymphoid aggregates and follicles, dense eosinophilic infiltration, and a few actinomycotic sulfur granules in the abscesses consistent with a diagnosis of actinomycosis (Fig.4). Therefore, she was treated with intravenous antibiotics for 2 weeks, followed by oral amoxicillin and clavulanate for 2 months.

The Institutional Review Board of 00 University Medical Center approved this case report and waived the requirement for informed consent (YUMC 2019-08-083).

**DISCUSSION**

Actinomycosis was first reported by Bradshaw in 1846 who described a patient with fistula formation between a right iliac fossa mass and the skin [3]. This condition commonly affects the oral cavity, the female genital tract, and the bronchial system. Therefore, actinomycosis is classified into cervicofacial, thoracic, and abdominopelvic types based on the site of involvement [4]. Abdominopelvic actinomycosis is one of the main clinical types and is commonly associated with a >4-year history of IUD implantation [5].
Clinical symptoms are characterized by contiguous spread, suppurative and granulomatous inflammation, formation of multiple abscesses and sinus tracts resulting in dense fibrosis and formation of a hard mass. Additionally, individuals with mucous membrane injury and immune deficiency are predisposed to actinomycosis. A. israelii (the main causative agent), becomes saprophytic under anaerobic conditions and penetrates the injured mucosa [6]. Invasion of host tissue precipitates an inflammatory response with the formation of multiloculated abscesses and pseudotumors.

Cope V.Z. reported that actinomycosis is one of the most misdiagnosed conditions and that no disease is so commonly missed even by skilled clinicians [3]. Preoperative diagnosis is challenging, particularly in patients with pelvic actinomycosis owing to the unusual presentation, low index of clinical suspicion, and difficulty with confirming actinomycetes as the causative organisms [7]. For example, actinomycosis of the GI tract or the parietal peritoneum is often indistinguishable from GI tract carcinoma or carcinomatosis. Notably, actinomycosis involving multiple sites of the parietal peritoneum is rare [6]. Although CT is a useful diagnostic modality for visualization of actinomycotic lesions, it cannot conclusively establish the diagnosis [8]. Radiologic findings are non-specific and are quite similar to those of Crohn’s disease, intestinal tuberculosis, or sometimes excavated malignant tumor [9]. Definitive diagnosis is necessarily based on isolation of actinomycetes from pus or histological examination of a suspicious mass. CT-guided aspiration with core needle biopsy is a useful diagnostic tool in cases of suspected actinomycosis. However, pus cultures rarely yield actinomycetes; therefore, these lesions are commonly misdiagnosed preoperatively as malignant tumors. Accurate diagnosis is only possible postoperatively based on histopathological examination of the surgically excised specimen [10].

The lesions in our patient were initially suspected to be malignant tumors rather than inflammatory lesions for several reasons. There were no signs of inflammatory responses such as fever or leukocytosis. Radiological findings on CT and MRI were consistent with multiple peritoneal masses, which were suggestive of malignant tumors rather than inflammatory abscesses. In addition, CA-125 was elevated in our patient.
Percutaneous needle biopsy was an option for tissue confirmation. However, we decided against percutaneous biopsy because small tissues obtained by needle biopsy, especially in cases of multiple huge mass similar to our case, cannot provide an accurate diagnosis. Surgical biopsy using laparoscopic exploration was not possible because of a huge mass in the peritoneal and pelvic cavity. We, therefore, performed exploratory laparotomy.

Treatment of abdominopelvic actinomycosis depends on the extent of the disease and the patient’s condition. Surgical resection is useful for definitive diagnosis and also as a therapeutic strategy for removal of infected tissue [11]. Antibiotic therapy alone can be considered in patients in whom actinomycosis can be conclusively diagnosed preoperatively. In our case, multiple intra-abdominal abscesses were identified, and carcinomatosis could not be ruled out. Therefore, surgical removal was performed, and actinomycosis was confirmed. Long-term antibiotic administration combined with surgical resection is warranted to ensure complete recovery from actinomycosis owing to inflammatory reactive fibrosis and the risk of relapse. Notably, *Actinomyces* spp. are usually susceptible to beta-lactams, particularly penicillin G. Clindamycin and tetracycline are useful alternatives in patients with adverse effects including penicillin allergy. The standard duration of antibiotic administration is unclear. In contrast to the previous recommended therapy of 6–12 months, short-term treatment over 2–6 weeks combined with surgical removal of lesions is known to be a successful treatment option [12].

We report a rare case of disseminated abdominopelvic actinomycosis mimicking carcinomatosis. Actinomycosis should be considered among the differential diagnosis in patients presenting with multiple abdominopelvic masses with radiological evidence of abscesses and a history of long-term IUD implantation.

**CONFLICT OF INTERESTS**

The authors declare no conflict of interests
REFERENCES


Figure legends

Fig. 1. Contrast-enhanced abdominal CT scan showing a huge enhancing mass connected to the greater omentum. A perirectal mass is also visualized.

CT: computed tomography

Fig. 2. PET scan showing a suspected carcinomatous lesion in the LLQ and multiple masses with positive FDG uptake

FDG: fluorodeoxyglucose, LLQ: left lower quadrant, PET: positron-emission tomography

Fig. 3. Histopathological examination showing gross findings. (a-b) A mass is visualized attached to the left lower parietal peritoneum and connected to the greater omentum. (c-d) A mass is visualized protruding from the right lower abdominal wall. (e-f) A perirectal mass is visualized.

Fig. 4. Histopathological examination showing microscopic findings. Diffuse inflammatory cell infiltration is visualized with multiple abscesses and fibrosis. An actinomycotic sulfur granule is visualized (inset). (Original magnification: main photo, ×20, inset, ×200)
Fig. 3