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Abdominal Actinomycosis Appearing as an Intraabdominal Tumoral Mass

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Introduction

Abdominal actinomycosis is an uncommon chronic infectious disease caused by a gram-positive anaerobic and microaerophilic bacterium, *Actinomyces israelii* (1). Clinical presentations of this entity are so variable that an accurate diagnosis cannot be established preoperatively in most cases. It can mimic inflammatory bowel disease, or even a malignant tumor (2,3). The clinical course of the disease is characterized by findings secondary to chronic inflammatory reaction and abscess formation. Delay in diagnosis makes the course worse and leads to abdominal septic complications (4). Recently, prolonged use of the intrauterine device (IUD) has been reported to increase the incidence of abdominal actinomycosis, and the gynecological tract is believed to be the most important source of infection (5). We report an unusual case of abdominal actinomycosis affecting the ileocecal region and gynecological organs and review literature experiences.

Case Report

A 29-year-old woman reported abdominal pain, nausea and vomiting. She had used an IUD for six years for contraception. On physical examination, the abdomen was diffusely tender and a vague tumoral mass was deeply palpated in the right iliac fossa. Routine blood tests were normal except for leukocytosis (13,500 cells/mm³) and mild anemia (Hb < 10 g/dl). Direct radiological examinations of the chest and abdomen showed no abnormality. However, abdominal ultrasonography and computed tomographic scans revealed a heterogenous mass 6 x 8 x 4.5-cm in size in the right iliac fossa (Fig. 1). Treatment was started with cephalosporin, but no improvement was achieved in

clinical appearance, and abdominal pain continued and worsened. She was therefore prepared for an operation, and a laparotomy was performed 1w after the initial treatment. Upon exploration, extensive granulomatous lesions involving the appendix vermiformis, the distal ileum and a small part of the omentum majus were encountered. There were also multiple abscess foci localized in the pericecal area. In addition to appendectomy, a segmentary resection of the distal ileum with primary anastomosis and excision of part of the omentum majus was carried out. When the abscess foci were entirely drained, the uterus, ovaries and tubes were left in situ. Aerobic culture was performed with a blood and eosin-methylene blue (EMB) agar at 37 °C for 24 h. Anaerobic culture was performed by using an anaerobic blood agar in Gas-Pak Jars at 37 °C for 24 h. However, no bacterial growth was observed within the cultures obtained from the abscess foci. The histopathological diagnosis of actinomycosis was confirmed by demonstrating the presence of sulfur granules, both in the segment of the ileum removed and in the portion of the omentum excised. On the histological section, a radial corona of eosinophilic material surrounded by bacterial masses of faintly visible branching filaments could be discerned (Fig. 2). Once a precise diagnosis had been established, the antibiotic regimen was altered to penicillin G, 10 million IU/day parenterally for 2 w, followed by oral therapy of 2 g/day for 2 months. The patient came back with recurrent abdominal pain 3 mon after the initial surgery. She underwent a comprehensive gynecological examination again. Pelvic ultrasound revealed a left tubo-ovarian abscess. On the second laparotomy, similar findings to those previously observed were encountered. Surgery was completed with a left

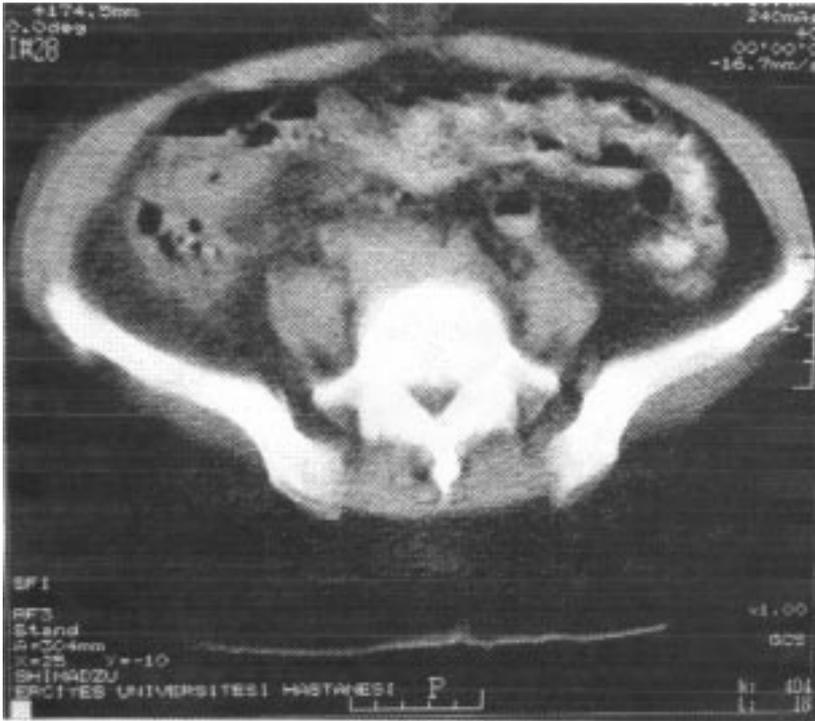


Figure 1. Computed tomographic scan showing a heterogeneous mass 6 x 8 x 4.5-cm in size in the right iliac fossa.

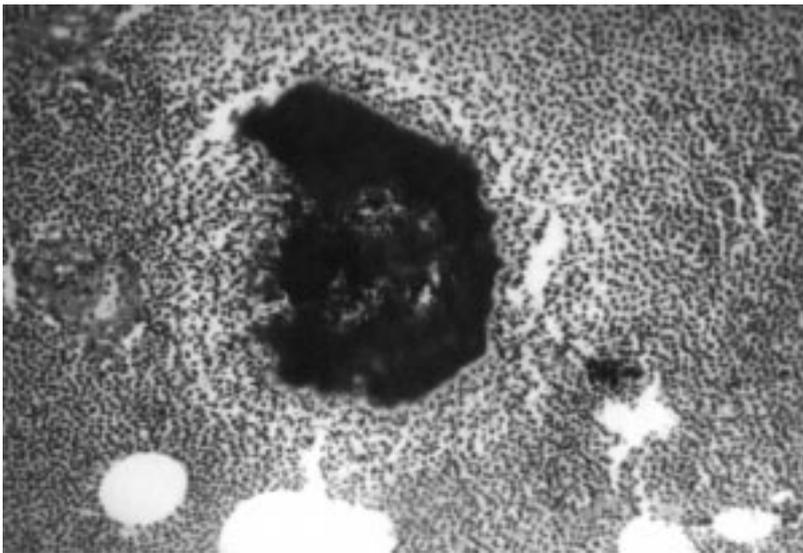


Figure 2. Histopathologic section shows characteristic sulfur granules produced by filamentous bacteria (H&E x 100).

salpingo-oophorectomy. Antibiotherapy of penicillin was continued for six months. The patient is doing well after one year of follow-up.

Abdominal actinomycosis is an uncommon chronic, suppurative infection caused by the gram-positive anaerobic and microaerophilic bacterium, *Actinomyces israelii* (1). Although the organisms are normal inhabitants of the oral cavity and gastrointestinal tract,

they rarely seem to be pathogens. In the early stage, the disease may be indistinguishable from other clinical conditions such as acute appendicitis, inflammatory bowel disease, acute cholecystitis or malignant tumor (2,3). The formation of multiple abscesses, sinuses and fistulas, fibrous adhesions and granulomatous lesions are all late-term signs (4,5). The ileocecal area is the most reported site for involvement (1-5). Other sites of involvement are

the colon, stomach, liver, biliary tract, spleen, kidneys and retroperitoneum (1-6). Recently, prolonged use of IUDs has been found to be related to pelvic actinomycosis (6,7). Our patient had been using an IUD for six years for contraception, although organisms could not be identified on vaginal examination. Nevertheless, we think that the patient's use of an IUD is a considerable risk factor.

Preoperative diagnosis of abdominal actinomycosis is difficult. No specific signs for this condition have been defined. Although radiological examination is not useful, a computed tomographic scan may be helpful in revealing an infiltrative mass with unusual aggressiveness and dense nonhomogeneous contrast enhancement (8). In our patient, computed tomography defined a heterogenous mass that was localized between the cecum and uterus, but we never considered this finding for preoperative diagnosis of actinomycosis (Fig. 1). Although the operative view was supportive, a definitive diagnosis of actinomycosis was made, indicating the sulfur granules in the resective materials after surgery (Fig. 2). While the presence of sulfur granules is considered to be an important finding, it is neither conclusive nor pathognomonic to *A. israelii*. Some other organisms, such as *nocardia*, *staphylococci* and *streptomyces*, may produce similar granules (2,4). Likewise, positive culture outcomes are also very helpful in the diagnosis of actinomycosis: however, this may be low in patients receiving penicillin because of the high sensitivity of *A. israelii* to this drug (9). Cultures from our patient from both surgical operations were negative for *A. israelii* and the other organisms. Initial use of broad spectrum

antibiotics may have influenced the culture yields. If no allergy exists, high doses and long-term use of penicillin is recommended to eradicate actinomycosis. It has a high capability of penetrating abscesses, sinuses and dense fibrotic structures, so it may be used as a therapeutic agent, alone or as an adjunct to surgery (9,10). Surgical treatment includes the excision of all necrotic tissues, drainage of abscesses and removal of sinuses and fistulas. More extensive surgery is rarely attempted (4,10). When recurrence occurs, repeated laparotomy may be required (1,9,11). We think that an inadequate duration of antibiotic cover led to the need for a second operation.

In conclusion, abdominal actinomycosis is an uncommon infectious disease that must be included among the differential diagnosis of infiltrating intraabdominal disorders. The use of IUDs for contraception remains an important risk factor. Accurate diagnosis is largely made with an examination of the specimen removed during surgery and demonstrating the presence of sulfur granules. Massive doses of penicillin are required for weeks to effect a cure in the majority of patients, but long-term use of antibiotics may be required in refractory instances to avoid a second laparotomy.

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