



Is appendectomy always adequate for treatment?: Clinical manifestations of isolated actinomycosis in the appendix

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ABSTRACT

Isolated appendiceal actinomycosis is a rare chronic progressive suppurative infection. Its causative agent in humans is a Gram-positive saprophytic anaerobic bacteria, *Actinomyces israelii*. We present a case of an acute appendicitis that developed in a 54-year-old woman due to isolated appendiceal actinomycosis. Diagnosis of appendiceal actinomycosis causing acute appendicitis is generally performed postoperatively histopathologically, and appendectomy alone is not sufficient for treatment. It is an important factor that should be considered by clinicians that definitive treatment of the infection is possible by appropriate antibiotic use.

Keywords: Actinomycosis, actinomyces, appendicitis, laparoscopic appendectomy

INTRODUCTION

Acute appendicitis, with an incidence of 7.6% (8.6% for males and 6.7% for females) in society, is the most commonly seen gastrointestinal surgical pathology that requires emergency surgery (1). It develops by the obstruction of the appendiceal lumen. Increased luminal pressure and impaired circulation cause bacterial propagation and increased mucus and induces the inflammatory response of the physiopathological process. The most frequent causes of obstruction are lymphoid hyperplasia, fecaloid, foreign materials, tumors, and parasites. Appendiceal actinomycosis is one of the rare causes of acute appendicitis.

Real acute actinomyces infection of the appendix can only be differentiated from ileocecal actinomycosis by actinomyces granules that are detected in the appendiceal lumen (2). It is an extremely rare case (2). Accurate diagnosis in those cases is clinically important since insufficient treatment may result in long-lasting disease risk, as frequently seen in local dissemination and metastatic abscess. On the other hand, chronic suppurative appendicitis is the clinical form that develops subsequent to the acute clinical picture (2).

We report a case of acute phlegmonous appendicitis caused by localized actinomyces infection alone.

CASE PRESENTATION

A 54-year-old female patient presented to our hospital with complaints of right lower abdominal pain, nausea, and vomiting for 2 days. Her medical history was unremarkable. On physical examination, her abdomen revealed rebound tenderness over the lower right abdominal quadrant, and no mass was palpable. A mild fever was noted at 37.9 °C. Laboratory findings were normal, except for white blood cell count of 14,500/mm³ and C-reactive protein level of 6.5 mg/L. Abdominal tomography without contrast enhancement revealed an enlarged proximal segment of the appendix (11 mm) and an increased wall thickness. Hyperdense material was detected to be compatible with appendicolith in the lumen. Computed tomography findings were compatible with acute appendicitis (Figure 1). Surgery was planned with suspected acute appendicitis. A laparoscopic appendectomy was performed. A vermiform appendix of 45 mm ×12 mm was found. Although the serosal layer of the appendix and mesoappendix was surrounded by fibrotic tissue, exploration of the ileocecal region was found to be normal (Figure 2). The patient recovered well postoperatively and was discharged from the hospital on postoperative day 1. Diagnosis of actinomycosis of the appendix was established histologically. Microscopic examination revealed acute phlegmonous appendicitis and actinomyces sulfur granules scattered in the purulent exudate in the appendiceal lumen (hematoxylin and eosin (H&E), ×2 magnification). A microscopic examination revealed typical sulfur granule in the lesion (H&E, ×10 magnification) (Figure 3). Actinomyces colonies were seen intramurally and intraluminally using Grocott's stain and Periodic acid-Schiff stain (Figure 4). After definitive diagnosis of appendiceal actinomycosis, the patient was readmitted to the hospital and treated with high doses of penicillin G intravenously for 2 weeks. Then, amoxicillin (2 g/day orally for 6 months) was prescribed to complete the treatment. Five years later, no clinical recurrence was noted.

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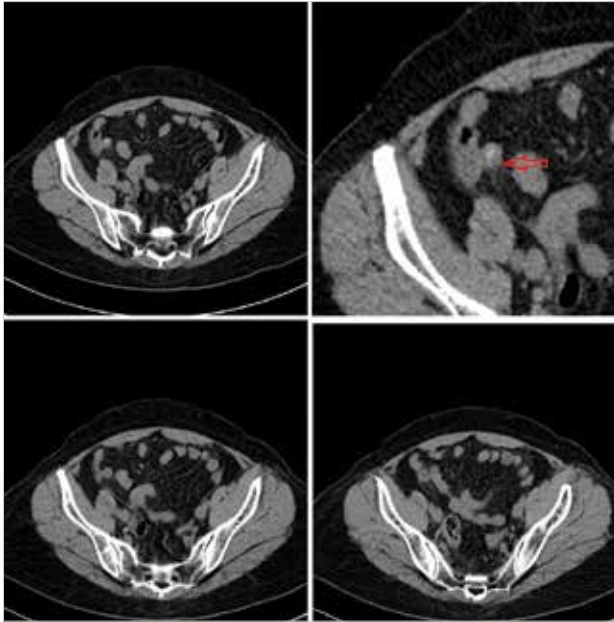


Figure 1. Abdominal computed tomography images. An enlarged proximal segment of the appendix (11 mm), increased wall thickness, and hyperdense material compatible with appendicolith in the lumen



Figure 2. Laparoscopic view. Serosal layer of the appendix and mesoappendix was surrounded by fibrotic tissue

Written informed consent was obtained from the patient who participated in the case study.

DISCUSSION

Actinomycosis is a subacute–chronic bacterial infection affecting the soft tissues and internal organs of the body. The most common pathogen that causes actinomycosis in humans is *Actinomyces israelii* (3, 4). This microorganism can be frequently found in the normal human mouth flora and less commonly in the lower gastrointestinal tract and female genital tract as well (4). Although *A. israelii* is a non-pathogenic bacteria, mucosal injuries that cause disruption of the mucosal barrier allow the microorganism to reach deep planes, resulting in actinomycosis infections in humans. The infection leads to the formation of granuloma and abscess, followed by consequent healing of the sinuses and drained fistulae. Involvement of distant or-

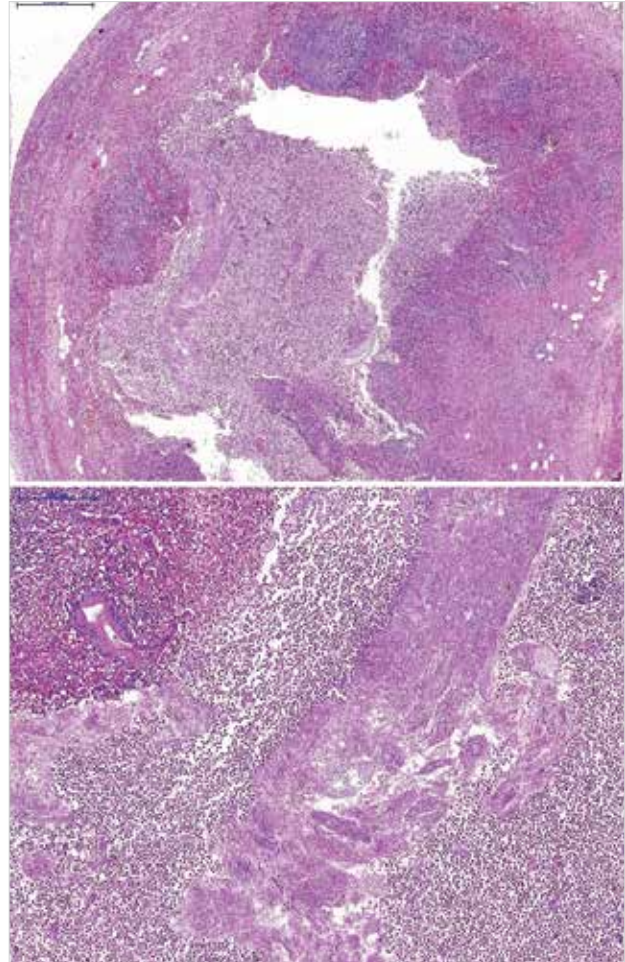


Figure 3. Diagnosis of actinomycosis was established with the presence of sulfur granules microscopically. Acute phlegmonous appendicitis. *Actinomyces* sulfur granules scattered in the purulent exudate in the appendiceal lumen (H&E, $\times 2$ magnification). *Actinomyces* sulfur granules (H&E, $\times 10$ magnification)

gans is possible via hematogenous metastasis at any stage of the disease (2). The disease may spread via the venous route, leading to the formation of metastatic abscesses in the liver. Actinomycosis cases among humans are seen in three forms, namely the cervicofacial form, which is the most frequent form observed in 50%–70% of cases, and less frequently, the thoracic and abdominopelvic forms observed in 15%–20% and 10%–20% of cases, respectively (3).

The disease is often seen in populations with low socioeconomic level and in individuals with bad oral hygiene aged between 20 and 50 years. Actinomycosis infection is more prevalent among men, with a male: female ratio of 3: 1, whereas the pelvic form is more frequent among women (3). The cause is often the prolonged use of intrauterine contraceptive devices, which cause chronic injury that allows the bacteria to penetrate the mucosal barrier and reach deep tissues (5, 6). The abdominal form of the disease is typically associated with a history of previous abdominal surgery. Pathologies that disrupt the integrity of the gastrointestinal mucosa, such as perforated appendicitis, perforated diverticulitis, endoscopic procedures, trauma, Crohn's disease, and intestinal perforation due to ingestion of bone or fishbone, are predisposing factors that play roles in the etiology of the disease (7).

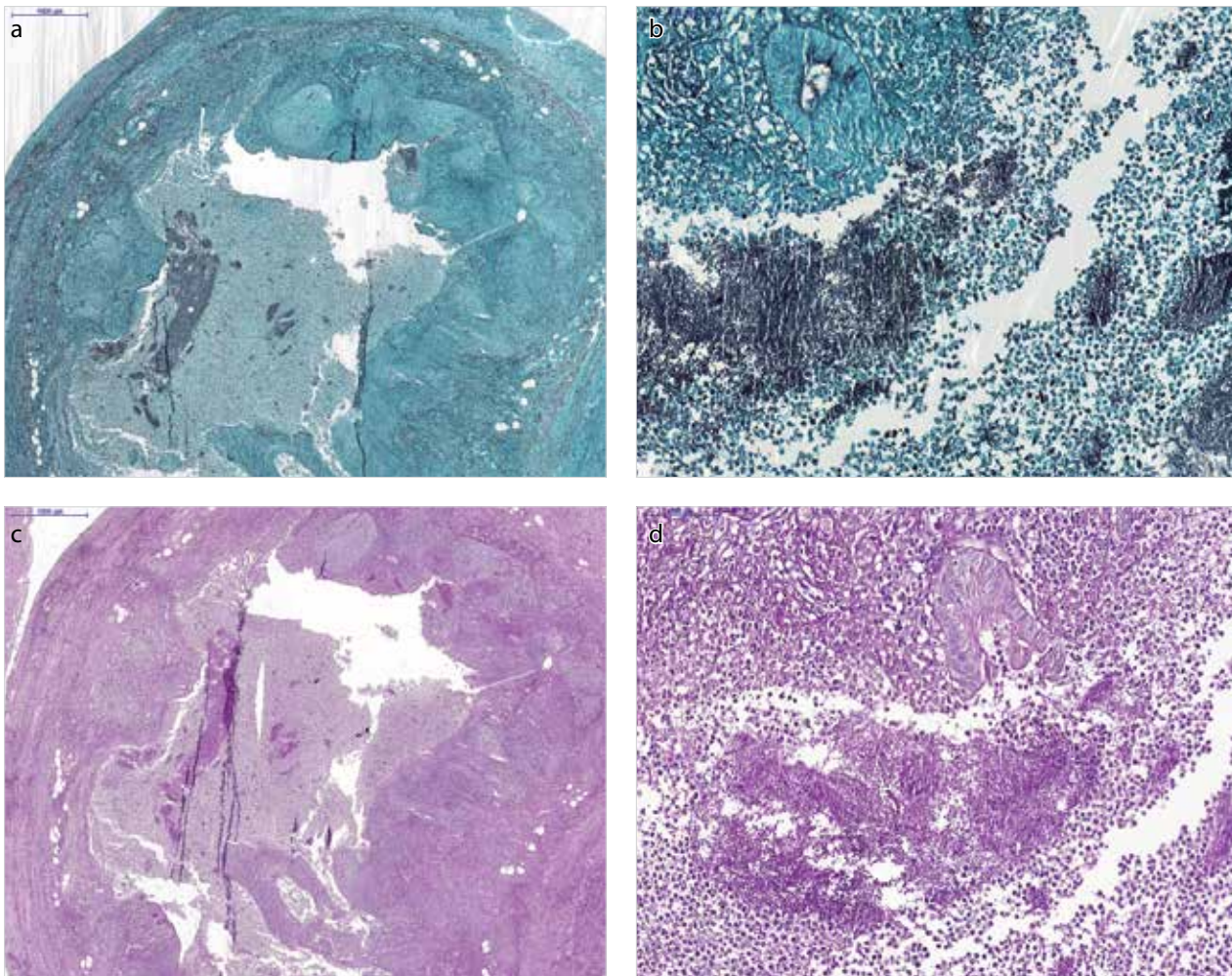


Figure 4. a-d. Colonies of actinomycetes (stained black with Grocott's stain) were seen intramurally and intraluminally. Actinomycetes sulfur granules positive with Grocott's methenamine silver stain ($\times 2$ magnification) (a), Grocott's methenamine silver stain ($\times 20$ magnification) (b), actinomycetes sulfur granules positive with Periodic acid-Schiff stain ($\times 2$ magnification) (c), periodic acid-Schiff stain ($\times 20$ magnification) (d)

Other known predisposing systemic factors include immunosuppression, malnutrition, and acquired immunodeficiency syndrome. Our case did not have any known local or systemic predisposing factor, such as history of previous abdominal surgery.

Abdominopelvic actinomycosis can be clinically manifested with fistula, sinus, inflammatory pseudotumor, or abscess, and it can often mimic other diseases clinically. The affected organ is surrounded by dense fibrous tissue, and the disease can mimic malignant diseases due to this mass appearance (3). Isolated actinomycosis of the appendix has non-specific clinical, laboratory, and radiological findings; therefore, the disease mimicking acute appendicitis is usually very difficult and often impossible to diagnose preoperatively (8). Radiological investigations are often non-diagnostic especially when there is no mass lesion or abscess concurrent with the diagnosis of acute appendicitis as in our case.

Diagnosis of abdominal actinomycosis could be made preoperatively in only 10% of the cases according to previous studies (6, 8, 9). Upon suspicion of the disease, diagnosis can be made with microscopical examination using a fine-needle aspiration material. However, diagnosis is often made with intraoperative or postoperative histopathological examination.

Histologically, the microorganism forms actinomycotic granules (sulfur granules) (10). These are made of irregular round bacterial aggregations surrounded by eosinophilic material, and they are typical for the disease. In our case, diagnosis was possible by observation of the typical sulfur granules detected during histopathological examination.

Studies report over 90% success rate in the treatment of abdominal actinomycosis infections when treated with a combination of antibiotics and surgical resection (6). The preferred antibiotic treatment includes high-dose crystalline penicillin G (2–4 weeks) and consequent long-term (6–12 months) oral penicillin or semi-synthetic penicillin derivatives (6, 7).

CONCLUSION

Appendiceal actinomycosis is a rare pathology that should be considered by clinicians to be among the causes of acute appendicitis. Accurate diagnosis is the key for successful treatment. Control and cure of the infection are only possible with long-term penicillin treatment after appendectomy.

Informed Consent: Written informed consent was obtained from the patient who participated in this study.

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