Journal of Pediatric Surgery (2007) **42**, E23–E26

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Journal of Pediatric Surgery

www.elsevier.com/locate/jpedsurg

Actinomycosis: a differential diagnosis for appendicitis A case report and review of the literature

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Index words:
Actinomyces;
Actinomycosis;
Appendix;
Children

Abstract *Actinomyces* is a genus of gram-positive anaerobic or microaerophilic bacteria that colonize the upper respiratory and gastrointestinal tracts and the female genital tract. These organisms cause disseminated disease in the mouth, the respiratory system, and rarely in the gastrointestinal tract. The diseases produced by *Actinomyces* species result from the disruption of the barriers that allow the dissemination of the bacteria through the surrounding tissues. The appendix is often a nidus of *Actinomyces* infection, but a prompt diagnosis cannot be made without the results of histologic examination of the appendix. The treatment of choice for actinomycosis of the appendix is the high-dose parenteral administration of penicillin G for 2 weeks immediately after the diagnosis has been made and continued oral treatment with that agent for at least the next 6 months. We present the case of a 13-year-old adolescent boy with actinomycosis of the appendix that was identified by histologic examination after appendectomy.

Actinomyces bacteria, which are found in the endogenous flora of the mucous membranes of the gastrointestinal, respiratory, and genital tracts, cause a disease referred to as *actinomycosis* (AMC), which develops when mucosal barriers are breached and those bacteria gain access to deeper tissues [1]. When compared with cervical and thoracic infections, abdominal AMC is a very rare disease that mimics several other disease processes and requires accurate diagnosis for successful treatment. The ileocecal region is the most frequent site for abdominal AMC [2]. That type of infection of the abdominal cavity is usually preceded by an obvious disruption of the mucosal barrier, such as that caused by abdominal surgery or appendicitis. However, cases without a known preceding incident have also been reported [3].

A diagnosis of AMC that is not based on the results of surgery or biopsy is almost inconceivable. Yellow or orange sulfur granules noted on histologic examination are a classic feature of AMC infection [4]. The treatment of AMC consists of the administration of high-dose intravenous penicillin G, followed by oral antibiotic therapy for at least the next 6 months [5]. We present the case of a 13-year-old adolescent boy with appendiceal AMC who was admitted with right lower abdominal pain to our institution. In the English literature, we have found only 12 reports of patients with abdominal AMC [6-17]. In this article, the rare occurrence of AMC in a child is discussed, and the English literature on that disorder is briefly reviewed.

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doi:10.1016/j.jpedsurg.2007.03.057

1. Case report

A 13-year-old adolescent boy with intermittent right lower abdominal pain and nausea of 4 weeks' duration was admitted to our institution. Physical examination revealed a palpable mass in the right lower abdominal quadrant but no signs of peritonitis. The results of urinalysis and the patient's whole blood count, blood chemistry values, and infection parameters were within the reference range. Abdominal ultrasonography and computed tomography revealed a thickness in the appendiceal wall, periappendiceal inflammation, and contrast enhancement. The patient underwent surgery for the removal of the appendiceal mass, at which time the appendix was found behind the cecum. The appendix was significantly swollen and was strongly adherent to the surrounding tissues; marked induration of the appendix was also noted. The appendix was excised. Subsequent macroscopic studies revealed no sign of gross perforation in the body of the appendix. Histologic examination showed sulfur granules and foci of *Actinomyces* colonization in the appendiceal lumen. The appendiceal wall was infiltrated with a fibrotic process, and a mononuclear inflammatory reaction with minor eosinophilic infiltration was noted. Muscular and serosal hypertrophy and clear lymphoid hyperplasia were evident, particularly in the distal appendiceal wall (Figs. 1 and 2). The diagnosis was AMC of the appendix vermiformis. The patient received high-dose (18 mU) penicillin G immediately after the diagnosis was established. Therapy with that agent was continued for the next 2 weeks, after which oral penicillin treatment was administered for the next 6 months.

2. Discussion

Actinomycosis is an indolent, slowly progressive infection caused by anaerobic or microaerophilic bacteria that are primarily from the genus *Actinomyces* and that usually colonize the mouth, colon, and vagina. *Actinomyces* species, which were originally classified as fungi because of their branching structures that are evident in culture and resemble hyphae, are true bacteria with neither mitochondria nor a nuclear membrane. Muramic acid is found in the cell wall of *Actinomyces* species, at least 14 of which have been identified to date. Most frequently, *Actinomyces israelii* and *Actinomyces naeslundii* have been the bacterial species isolated [1].

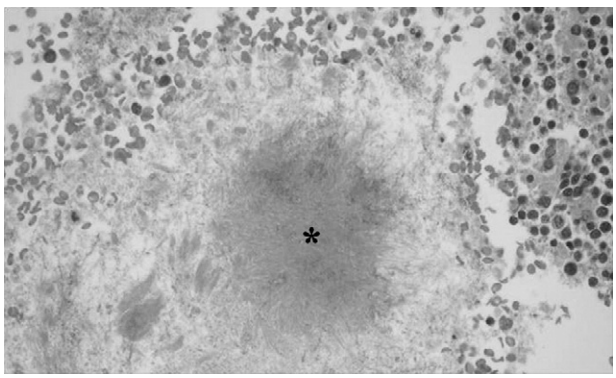

Fig. 1 *Actinomyces* colonies (*) (H&E stain, original magnification ×400).

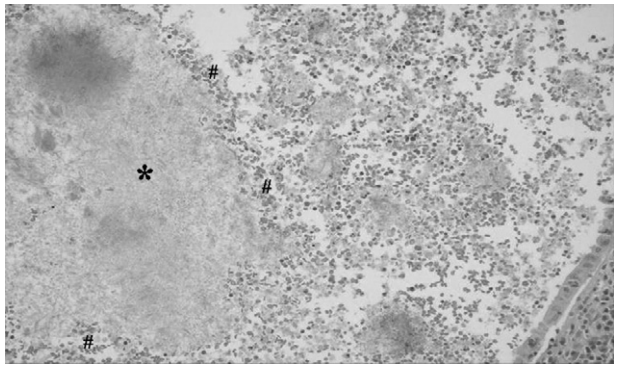

Fig. 2 *Actinomyces* colonies (*) and surrounding sulfur granules (#) (H&E stain, original magnification ×400).

A pivotal step in the pathogenesis of AMC is the disruption of the mucosal barrier [1,5]. Actinomycosis is most frequently manifested as oral or cervicofacial disease. The lung is the next most common organ affected by AMC, which often develops after aspiration of the bacteria by the host [18]. Manifestations of the disease include persistent pneumonia and pulmonary infiltrates or a lesion discovered on routine chest radiographs. In untreated or disseminated cases, AMC may spread to the retroperitoneum through the diaphragmatic sinuses [11]. Abdominal AMC was first described in 1949 by the English surgeon Bradshaw as a mass in the right iliac fossa [19]. The peak incidence of AMC is reported to occur in middle-aged individuals; cases in people younger than 10 years or older than 60 years occur less frequently [1]. In children, abdominal infection caused by *Actinomyces* bacteria is rare. In a review of the English literature, we identified only 12 reports of children with abdominal AMC [6-17].

In patients with visceral AMC, the appendiceal area is by far the most common site affected. The infection less frequently involves the colon, stomach, liver, gallbladder, pancreas, small intestine, anorectal area, pelvis, or abdominal wall [2]. Although retroperitoneal disease has been reported to be more frequent than the infection of intraperitoneal organs [11], our review indicated that about half the patients with AMC also had appendiceal or liver involvement. In most patients with AMC, a single organ is infected; the disseminated form is rare. The inflammatory process of AMC is divided into 3 phases. In the first phase, the abscess is limited to its original site in the parenchyma, and clinical manifestations may be absent. In the second phase, the peritoneum is involved either as a result of a localized abscess or more often by the dissemination of the infection. In the third phase, fistulas form.