CASE REPORT

P. Schmidt · J. L. Koltai · A. Weltzien

Actinomycosis of the appendix in childhood

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Abstract Abdominal actinomycosis (AMC) is a rare infection in children. The appendix is the most common intra-abdominal organ involved. It presents as an undifferentiated mass, forming abscesses and fistulas in the right lower quadrant. The case of a 15-year-old girl with a AMC of the appendix detected by the pathologist after routine appendectomy is discussed. Long-term antibiotic treatment and follow-up by ultrasound and laboratory controls are necessary.

Key words Actinomycosis · Antibiotic treatment · Appendicitis · Children

Introduction

Actinomycosis (AMC) is a chronic and progressive infection of soft tissue and viscera causing fibrosis and suppuration. If forms abscesses, fistulas, and abundant granulation, and the tissue becomes extremely tough. The appearance of sulfur granules in the lesions is a histologic characteristic [4, 10]. The most important species in humans is Actinomyces israelii, which was first described by Israel in 1878 [11]. This anaerobic, gram-positive bacterium is a commensal of the oropharynx, and is also found less frequently in other sites of the intestinal tract [4]. It remains a matter of discussion whether the organism can penetrate the intact mucous membrane and produce the primary lesion by itself, or if infection only occurs if the normal barrier of intact gastrointestinal mucosa has been destroyed by infection or trauma [10]. AMC develops mainly in the soft tissue of the face, neck, and oral cavity. In comparison to cervical and thoracic forms, abdominal infection is quite rare [3, 15, 20, 26, 27].

The case of a 15-year-old girl is discussed; she presented with severe abdominal pain. At surgical intervention, appendicitis with massive swelling of the appendiceal wall was found. Histologically, AMC of the appendix was detected.

Case report

A 15-year-old girl suffered from abdominal pain for 2 days, with nausea but no vomiting or diarrhea. There was right lower quadrant (RLQ) pain but no sign of peritonitis. There was a palpable mass in the RLQ. The white blood cell count was 11,200/μl and serum C-reactive protein was markedly increased at 9.8 mg/dl. Abdominal ultrasound (US) showed a target sign in the RLQ measuring 2.5 cm on cross-section. This was interpreted as an intussusception. A contrast enema appeared to confirm their diagnosis. At surgery, the appendix was enlarged without signs of perforation (Fig. 1). The appendiceal wall protruded into the cecum like an intussusception. The walls of the appendix and cecum showed marked induration. A small amount of pus was found in the appendiceal lumen; a swab was taken for microbiologic examination. The appendix and cecal pole were resected.

Macroscopic examination (Fig. 2) revealed a massively swollen but not destroyed appendiceal wall. Microscopic examination (Fig. 3) showed typical sulfur granules and small, superficial sinuses in the appendiceal lumen. Theecal wall was involved in the fibrotic process and showed suppurrative infection. The culture showed superinfection with Bacteroides fragilis. Postoperative antibiotic treatment consisted of high doses of penicillin IV for 4 weeks. Because of an allergic reaction, this was changed to oral tetracycline. Treatment was continued for 1 year. Eight months after the operation, there had been no complications. The diagnosis of AMC of the appendix was established histologically.

Discussion

The English surgeon Bradshaw was the first to describe abdominal AMC, which was found in the right iliac fossa [5]. Most publications since that time have been case reports [1, 12, 14, 19, 26, 28]. In childhood, abdominal infections caused by Actinomyces are rare [1, 7, 16, 22]. Retroperitoneal disease is more frequent than infection of intraperitoneal organs [22]. Infection of the intraperitoneum is thought to be caused by ascending bacteria [25]. Intraperitoneal bacterial dissemination seems to originate mainly from the appendix [16, 24]. The vermiform

P. Schmidt (✉) · J.L. Koltai · A. Weltzien
Kinderchirurgie, Städtische Kliniken Frankfurt/M.-Höchst, Gothenstrasse 6-8, D-65929 Frankfurt am Main-Höchst, Germany
appendix is by far the most common intra-abdominal organ involved [2]. The correct diagnosis is usually not established before operation [8,9,29]. Often the disease is only discovered after infection spreads within the peritoneal cavity or when percutaneous fistulas become visible [1].

The typical presentation of abdominal AMC as an inflammatory pseudotumor in the RLQ is often missed [2,3,22]. The diagnosis is even missed by US, radiographic, or computed tomographic examinations [17,22]. A needle biopsy is only helpful if sulfur granules are found [6,12,24]. In microbiologic studies Actinomyces is difficult to isolate by culture because the bacteria need very special conditions for growth [4,10]. In most cases the diagnosis is established by the pathologist [8,9,20,29]. Abdominal AMC most frequently presents an undifferentiated mass, forming abscesses and sinuses in the RLQ [22]. The infection spreads by continuity from one organ to the next. At operations for suspected appendicitis, the appendix infected by Actinomyces shows typical sinuses [9,20,21,24]. There are only a few descriptions of an intact appendix in cases of AMC [21]. If the diagnosis is missed, AMC can occur after appendectomy [2,3], but after routine appendectomy for appendicitis this is very rare [13].

If the diagnosis can be made without surgery, the treatment of choice is with antibiotics. Abscesses must be drained and sinuses excised [8,14,15,29]. If the diagnosis is made at surgery, appropriate treatment with high doses of antibiotics should follow. Although Penicillin is the drug of choice, tetracycline, lincomycin, and clindamycin are suitable alternatives in the penicillin-allergic patient [3,4,18], administered parenterally for 2 to 4 weeks and followed by prolonged oral therapy of 6 to 12 months’ duration [9,10,23]. Laboratory and US examinations should be done every 4 weeks. If this regime is followed, AMC has a favorable prognosis [2–4,7,10,15,16,18,23,27]. If inadequate postoperative antibiotic treatment is used, the infection will relapse [1,21,23]. Prior to the antibiotic era, the outcome of abdominal AMC was fatal [2,12,24].

As this case demonstrates, abdominal AMC in childhood remains a rare and problematic infection even if it is found incidentally during an appendectomy.