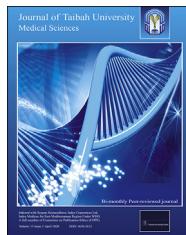




Taibah University

Journal of Taibah University Medical Sciences

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Case Report

Caecum actinomycosis with acute abdomen: A case report

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Received 19 November 2019; revised 19 January 2020; accepted 20 January 2020; Available online 19 March 2020

الملخص

داء الشعوب البطني، أحد أسباب اضطرابات اللانفاني الأعورى، وعادةً ما يتم التفكير فيه عند استبعاد الحالات السريرية الأخرى الأكثر شيوعاً. داء الشعوب هو اضطراب جرثومي نادر ينجم عن أنواع الإكتينوميسيات. نقدم جندياً سعودياً يبلغ من العمر ٣٨ عاماً تعرض لآلم في الحفرة الهرقانية اليمنى لأربعة أيام. بدأ هذا الألم على شكل طعن تدريجياً. بناءً على الفحص السريري للمريض والموجات فوق الصوتية للطن، تم إجراء عملية استئصال الزائدة الدودية. وأظهر الفحص النسيجي داء الشعوب الزاندي مع تضخم اللقافية، والاحتقان المصلي والبكتيريا الخبيثة في تحريف الزائدة الدودية. تم علاج المريض بالأموكسيسين. وأظهرت الأشعة المقطعيّة مع الصبغة للطن، وأظهر التصوير بالرنين المغناطيسي ارتفاع سماكة جدار الأعور 4.3×2.9 سم. خضع المريض أخيراً لاستئصال اللانفاني الأعورى بمساعدة التقطير مع مغاغرة اللانفاني القولوني. أظهر التقرير النسيجي مواد غذائية متخلسة في الرتج مع التهاب مزمن دون داء الشعوب التي كان يمكن علاجها والقضاء عليها من خلال العلاج بالمضادات الحيوية السابقة.

الكلمات المفتاحية: البطن الحاد؛ وجع بطن؛ داء الشعوب؛ استئصال الزائدة الدودية؛ استئصال اللانفانية

Abstract

Abdominal actinomycosis, one of the causes of ileocaecal disorders, is usually considered when other more common

clinical conditions have been excluded. Actinomycosis is a rare infectious bacterial disorder caused by the *Actinomyces* species. We present the case of a 38-year male Saudi soldier who presented with pain in the right iliac fossa since 4 days prior to presentation. This stabbing pain started gradually. Based on clinical examination and abdominal ultrasound findings, an appendectomy was performed. Histological examination revealed appendicular actinomycosis with lymphoid hyperplasia, serosa congestion, and filamentous bacteria in the appendicular lumen. The patient was treated with amoxicillin. During follow-up, contrast-enhanced abdominal computed tomography (CT) and magnetic resonance imaging (MRI) revealed a 4.3×2.9 cm thickened caecal wall. Thereafter, the patient underwent laparoscope-assisted ileocaecal resection with ileocolic anastomosis. The histological report revealed calcified food material in the diverticulum, with chronic inflammation without actinomycosis, which may have been eradicated by the previous antibiotic treatment.

Keywords: Acute abdomen; Abdominal pain; Actinomycosis; Appendectomy; Caecum actinomycosis; Ileocaecal resection

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Introduction

According to general surgical services, right iliac fossa (RIF) pain is a common condition.¹ The differential

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Peer review under responsibility of Taibah University.



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diagnosis is widely varied and ranges from appendicitis to urological, gynaecological, vascular, and musculoskeletal disorders. Because clinical presentation in all these is analogous, the true cause may be uncertain, especially in case of ovarian pathologies in females of reproductive age. Therefore, diagnosing appendicitis can be challenging.^{2,3}

Abdominal actinomycosis is one of the causes of ileo-caecal lesions. It is generally deliberated in specific clinical settings or when the more common causes have been excluded or are improbable.⁴ Actinomycosis is an uncommon, infectious bacterial disease caused by the *Actinomyces* species.⁵ About 70% of the infections are due to either *Actinomyces israelii* or *Actinomyces gerencseriae*.⁶ Infection can also be caused by other *Actinomyces* species, as well as by *Propionibacterium propionicus*, which presents similar symptoms. The condition is likely to be a polymicrobial aerobic/anaerobic infection.⁶

Case report

A 38-year-old male Saudi soldier from a rural area presented to our hospital with RIF pain that began four days ago. He had no relevant medical history. His symptoms started gradually as a localized stabbing pain that was aggravated by movement and decreased with rest. One day prior to presentation, the pain was associated with vomiting (no blood). An examination revealed that the patient was afebrile. The body temperature was 36.1 °C and the blood pressure was 110/78 mmHg. On palpation, the abdomen was observed to be soft and lax with no tenderness at the RIF. Abdominal ultrasound revealed free minimal fluid on the RIF, with echogenic fat planes at the assumed location of the appendix. Furthermore, a tubular structure having a maximum transverse diameter of 5.4 cm was also detected. Laboratory findings were as follows: white blood cell: 8.7, red blood cell: 4.9, haemoglobin: 15.2, platelet count: 316, total bilirubin: 22.5, direct bilirubin: 5.5, alanine aminotransferase: 27U/L, aspartate aminotransferase: 30U/L,



Figure 1: CECT shows marked thickening of the caecum (short arrow) with a narrowed lumen, and to a lesser extent, the terminal ileum (long arrow) surrounding the fat stranding.



Figure 2: CT scan of the lower abdomen shows a dense foreign body (long arrow) impeded within the caecal wall and the fat strandings (short arrow).

alkaline phosphatase: 48U/L, gamma-glutamyl transferase: 16U/L, C-reactive protein: 23.8mg/l, creatinine: 80 µmol/L, and blood urea nitrogen: 4.8µmol/L. An appendectomy was performed, and subsequent histopathology revealed lymphoid hyperplasia with serosa congestion and filamentous bacteria in the lumen (Actinomycosis). The patient



Figure 3: Coronal reformatted CT shows the foreign body with surrounding hypodensity representing collections, caecal wall thickening (short arrow), and fat stranding (long heads). Based on these findings, actinomycosis infection is suggested.

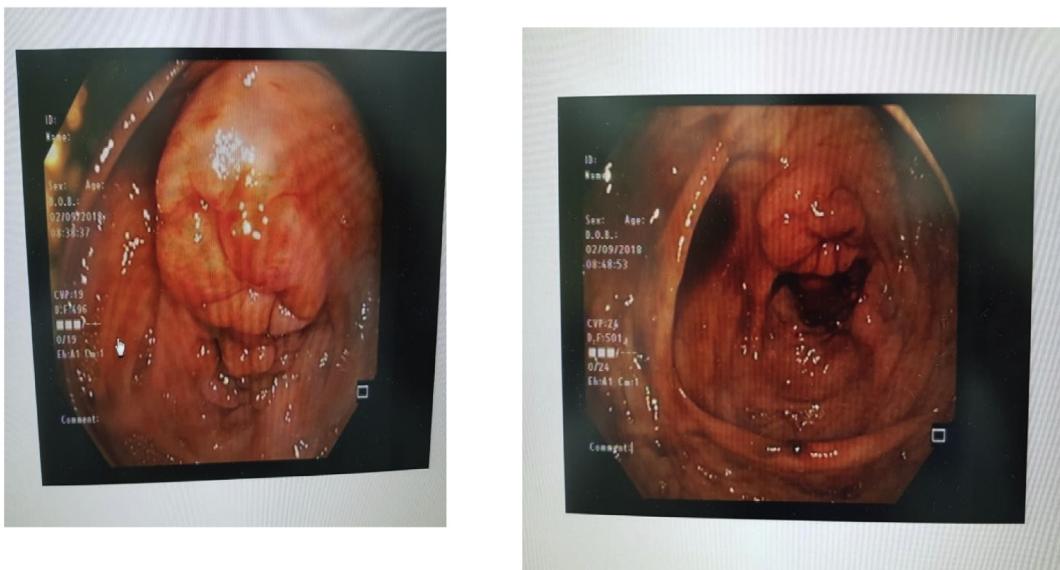


Figure 4: Colonoscopy reveals a polyloid lesion at the base of the caecum involving the appendix opening, hard on biopsy.

underwent abdominal contrast-enhanced computed tomography (CECT) and was started on amoxicillin. CT revealed extensive wall thickening in the distal ileum along with surrounding fat (Figures 1–3). One month later, while the patient was still on amoxicillin, a follow-up abdominal CT scan revealed relative regression of the wall thickening that involved the distal ileum and caecum. However, the patient still complained of abdominal pain that recurred at least once a week. Thus, he was scheduled for contrast-enhanced abdominal magnetic resonance imaging (MRI), which revealed a caecal mass with wall thickening, measuring about 4.3×2.9 cm. Colonoscopy revealed a polypliod mass at the appendix (caecal mass, Figure 4). A biopsy was taken and sent for histopathology and culture. Histopathology revealed a colonic mucosal fragment with ill-defined

granulomatous reaction and prominent eosinophilia with no dysplasia. Fungus culture did not yield an isolate. The patient then underwent a laparoscope-assisted ileocecal resection with ileocolic anastomosis. No postoperative complications were observed (Figure 5) and histopathology report calcified food material in diverticulosis with chronic inflammation.

Discussion

Actinomycosis is an uncommon, chronic granulomatous disease caused by filamentous, gram-positive, anaerobic bacteria.⁷ *A. israelii* is the main causative agent in humans.^{1,2} Actinomycosis has a global distribution. It mainly affects the middle-aged populations and is two to four times more common in males.^{8–10} Actinomycetes are the normal inhabitants of the oral cavity and the gut⁵; however, they develop pathogenicity upon invasion of breached or necrotic tissue. As the infection progresses, granulomatous tissue, massive reactive fibrosis and necrosis, abscesses, draining sinuses, and fistulas are formed.⁷ The cervicofacial area is the most frequently infected (50%), followed by the abdominal area (20%), and the thoracic area (15%–20%).¹¹ In abdominal actinomycosis, the appendix and the ileocaecal region are usually involved. The infection mostly remains localized; it then spreads contiguously, disregarding tissue planes. Lymphadenopathy is not a common finding. Hematogenous dissemination is also rare.⁸ The causative agents are the normal inhabitants of the mucous lining in the nose, throat, mouth, intestinal tract, and the female reproductive system, and are not naturally harmful. These anaerobic bacteria have the ability to grow in either the absence or in reduced concentrations of oxygen. However, any injury, trauma, or surgical procedure can cause the bacterial cells to enter deeper tissues, where they normally do not exist. Because these bacteria can grow without oxygen, they can thrive in such environments, resulting in infection.¹²

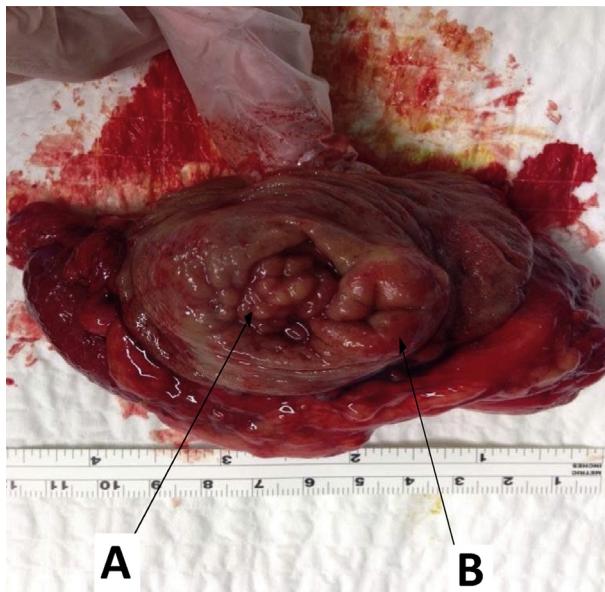


Figure 5: (A) Shows a lesion at the site of the appendiceal orifice. (B) Ileocaecal junction.

Intestinal tract actinomycosis is a rare, chronic bacterial infection of the abdominal mucosa and/or organs of the digestive tract, mainly caused by the bacterium *A. israelii*, and sometimes by other *Actinomyces* species.¹³ Actinomycosis of the caecum is a rare, chronic bacterial infection of a part of the large intestine, caused predominantly by the bacterium *A. israelii*, and to a lesser extent by other *Actinomyces* species.¹⁴ The diagnosis of abdominal actinomycosis is challenging and needs surgical assessment through intervention. The cases usually have vague, nonspecific clinical complaints and the most frequent symptom is abdominal pain corresponding to the site of the infected organ.^{15,16} The course of the disease is indolent and is similar to that of other diseases such as appendicitis, diverticulitis, colon carcinoma, Crohn's disease, ulcerative colitis, and tubo-ovarian abscess.¹⁷ In the early stages, the disease is often confused with appendicitis, carcinoma caecum, tuberculosis, or amoebiasis. The preoperative diagnosis may depend mainly on the findings of imaging modalities, which often cannot discriminate between actinomycosis and malignant process, Crohn's disease, appendicitis, diverticulitis, or tuberculosis^{18,19}; however, a majority of the cases can be confirmed after surgery, based on macroscopic, microscopic, and histochemical examinations of the specimen after surgical exploration.¹⁸

Evidence increasingly indicates that medical therapy alone, without surgical exploration, is the main line of treatment, irrespective of the degree or severity of the infection.^{16,20} Treatment of actinomycosis consists of intravenous Penicillin-G for four weeks, followed by oral penicillin for six to 12 months.^{21,22} Although no true surgical intervention guidelines have been established, operative treatment has been pursued in patients who present with extensive necrotic tissue or large abscesses that cannot be adequately drained.²³

Our patient continued to receive amoxicillin till definitive pathology. No complications arising from the surgical excision and anastomosis were observed during the follow-up. The histopathology showed no actinomycosis, which may indicate that the patient responded to the antibiotic treatment.

Source of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Conflict of interest

There is no conflict of interest.

Ethical approval

The case report was approved by research ethics committee at July 15, 2019 with code No. 372.

Authors contributions

BIA and EAA conceived, collected, organized, and designed the study, participated in writing the initial and

final draft of the article. AAA participated in writing the final draft and critically revised the manuscript for intellectual content. ASA, AMA, and YIA provided the pictures and participated in writing part of the initial draft. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

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How to cite this article: Asiri BI, Alshehri AA, Alqahtani AS, Albishi AM, Assiri YI, Asmiri EA. Caecum actinomycosis with acute abdomen: A case report. *J Taibah Univ Med Sc* 2020;15(2):148–152.