



CLINICAL CASE

Hepatic Actinomycosis



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Abstract

Actinomycosis is a rare disorder caused by an anaerobic gram-positive bacillus (*Actinomyces*), predominantly by the *Actinomyces israelii* species. Only 20% of cases show an abdominal manifestation, the appendix and ileocecal valve being the most frequent locations. Definitive diagnosis is based on microbiological cultures, microscopy or macroscopy examination. Nevertheless, histological examination of the percutaneous biopsy and blood microbiological cultures are rarely positives. Preoperative diagnosis is hampered by the lack of specific clinical and imaging manifestations, which often mimic malignancy. The rate of preoperative diagnosis is less than 10%, however, the outcome is excellent, with a low mortality rate. The authors describe the case of a patient who was diagnosed with primary hepatic actinomycosis only by a histological examination of the surgical specimen of left hepatectomy extended to segments V and VIII, for suspected malignant lesion. This case demonstrates the difficulties in diagnosing hepatic actinomycosis.

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PALAVRAS-CHAVE

Actinomyces;
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Doença
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Doenças do Fígado

Actinomicose Hepática

Resumo

A actinomicose é uma entidade clínica rara, causada por uma bactéria anaeróbia gram-positiva (*Actinomyces*), predominantemente da espécie *Actinomyces israelii*. Apenas em 20% dos casos apresenta manifestação abdominal, sendo o apêndice e a válvula ileocecal as

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localizações mais frequentes. Os autores descrevem o caso de um doente em que foi feito o diagnóstico de actinomicose hepática primária apenas pelo exame histológico da peça cirúrgica de hepatectomia esquerda alargada aos segmentos V e VIII, por suspeita de lesão maligna. Este caso demonstra a dificuldade diagnóstica da actinomicose hepática. O diagnóstico pré-operatório é dificultado pela falta de manifestações clínicas e imagiológicas específicas, muitas vezes simulando doença maligna. Para além disso, as culturas e o exame histológico de biópsia percutânea raramente são positivos. A taxa de diagnóstico pré-operatório é inferior a 10%, contudo o prognóstico é bom, apresentando uma taxa de mortalidade de cerca de 7,6%.

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1. Introduction

Actinomycosis is a rare chronic, granulomatous infection caused by gram-positive bacilli of the genus *Actinomyces*. There are 13 different species, but only 6 are associated with human disease. The most common pathogenic specie in humans is *Actinomyces israeli*.¹ These microorganisms are prominent among the commensal flora of the oropharynx, gastrointestinal tract and female genital tract.² Actinomycosis is most frequently associated with cervicofacial infection. Only 20% of the cases present abdominal manifestations, the appendix and the ileocecal region being the most common affected sites.³ Liver involvement occurs in 15% of abdominal infections representing 5% of all cases of actinomycosis,⁴ and it is usually secondary to other intraabdominal infection.⁵ The insidious course with nonspecific symptoms makes the diagnosis of this condition a challenging.

2. Case report

A 38-year-old man was referred to the Gastroenterology Department for a history of epigastric pain refractory to proton pump inhibitor therapy. He reported a 2 years history of night sweats and weight loss (12% of body weight). The patient denied other dyspeptic complaints, hematemesis or melena and systemic symptoms, including fever, malaise and anorexia. His past medical history included duodenal ulcer treated with proton pump inhibitor, 3 years ago. No history of previous surgeries. He was medicated with omeprazole 20mg qd on regular basis. The physical examination was normal. Laboratory tests evidenced normocytic/normochromic anemia (Hb 12 g/dL, normal: 14–18), leukocytosis ($15.21 \times 10^3/\mu\text{L}$, normal: 4–11.5) with neutrophilia (80%, normal: 50–70), elevation of inflammatory parameters (C-reactive protein: 2.46 mg/dL (normal 0–0.3) and erythrocyte sedimentation rate 33 mm (normal 0–15) and mild elevation of alkaline phosphatase 170 U/L (normal: 40–129). The remaining analytical assessment was normal, including tumor markers and human immunodeficiency virus antibody test. Infection by *Mycobacterium tuberculosis* was excluded. Upper gastrointestinal endoscopy revealed pyloric substenosis, yet allowing the passage of the endoscope

to the duodenum, probably resulting from previous duodenal ulcer. Biopsies were performed in normal mucosa, which revealed chronic gastritis due to *Helicobacter pylori*. Gastroduodenal transit was normal. An abdominal ultrasound identified in segment IV of the liver a pseudonodular lesion, hypoechoic, heterogeneous with partially undefined limits with 45×33 mm (Fig. 1). A contrast-enhanced computed tomography (CT) scan showed the same lesion, presenting low enhanced in the arterial phase, with progressive enhanced in the late phase (Fig. 2). To further characterize the lesion a magnetic resonance imaging (MRI) was performed revealing the same nodular lesion with lobulated and undefined limits of 4 cm in its greatest diameter, hypointense signal on T1 and hyperintense signal on T2 (Fig. 3). These images were suggestive of cholangiocarcinoma but secondary deposit cannot be excluded. A CT-guided liver biopsy revealed an inflammatory pseudotumor/inflammatory myofibroblast tumor (Fig. 4). The patient was then referred to referral Hospital in hepatobiliary surgery where he underwent left hepatectomy extended to segments V and VIII. The histological examination of surgical tissue demonstrated abscesses due to *Actinomyces* (Fig. 5). The patient was treated with doxycycline for three months. Eighteen months after the surgery, he is completely asymptomatic.

3. Discussion

Actinomycosis is a rare cause of intra-abdominal infection. The main risk factors associated with this condition are loss of integrity of the gastrointestinal mucosa, previous abdominal surgery, intra-abdominal infection, gastrointestinal foreign body and immunosuppression.^{5,6} Liver involvement occurs by hematogenous spread via the portal vein from either a mucosal injury or infection.⁷ Sharma et al. conducted a study which found that about 17.5% of cases of abdominal actinomycosis were associated with previous abdominal or pelvic procedures performed within the last year.⁸ In this case, the time between diagnosis and the initiation of duodenal ulcer was one year. Despite the known major risk factors, hepatic actinomycosis is cryptogenic in 80% of cases and^{4,6} polymicrobial in 33%.⁹ As in our case, there is male predominance (70–74%) in immunocompetent

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