

Chronic typhlitis in a non-neutropenic patient associated with actinomyces. Case presentation

Tiflitis crónica en paciente no neutropénica asociado a *Actinomyces*. Presentación de caso

Juan Pablo Manrique Salas[1]

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Palabras clave (DeCS)

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Summary

Actinomycosis is a chronic suppurative disease caused by Actinomyces species, which are members of the endogenous flora normally found in the mouth, gastrointestinal tract, and female genital tract. The clinical and radiological findings of actinomycosis are variable, depending on the primary site of involvement and the duration of the disease. In fact, it can often resemble other inflammatory and neoplastic lesions, making actinomycosis a diagnostic challenge. The case of an 8-year-old female patient who presents with abdominal pain and mass is presented, evaluating the ultrasound and tomographic images in comparison with the findings at surgery.

Resumen

La actinomicosis es una enfermedad supurativa crónica causada por especies de *Actinomyces,* miembros de la flora endógena, que normalmente se encuentran en la boca, el tracto gastrointestinal y el tracto genital femenino. Los hallazgos clínicos y radiológicos de la actinomicosis son variables, según el sitio primario de afectación y la duración de la enfermedad. De hecho, con frecuencia puede parecerse a otras lesiones inflamatorias y neoplásicas, por lo que la actinomicosis plantea un desafío diagnóstico. Se presenta el caso de una paciente de 8 años de edad, quien cursa con dolor y masa abdominal; se evalúan las imágenes ecográficas y tomográficas y se comparan con los hallazgos en la cirugía.

Introduction

Actinomyces infection is an infrequent and chronic inflammatory process that can simulate an inflammatory bowel disease, diverticulitis, tuberculosis or even a malignant tumor; for this reason, it can also be called an inflammatory pseudotumor. This pathology may require surgical treatment when it cannot be distinguished from other diseases or due to intestinal complications (1).

Preoperative clinical and radiological diagnosis is rarely performed. The clinical picture of abdominal involvement has nonspecific signs such as fever, weight loss, constipation or diarrhea, nausea, vomiting, anorexia and abdominal pain, in addition to abdominal mass and anemia. Occasionally there is leukocytosis in the blood picture. Radiologically, actinomycosis masses on computed tomography (CT) most often present as predominantly solid lesions or as mixed cystic and solid lesions, with blurred margins and irregularly thickened walls. In addition, CT allows a more precise definition of the dimensions and extent of the infection. This same diagnostic method can be used as a guide for percutaneous biopsy and abscess drainage, which would help in the preoperative diagnosis (2).

Case report

An 8-year-old female patient with a history of diabetes mellitus and hypothyroidism, who presented with abdominal pain of six months of evolution, located in the right flank, characterized by cramping without triggers. No fever, weight loss, diarrhea or vomiting. She manifests sensation of increased volume in the periumbilical region, extending to the right iliac fossa, which has been progressive and accompanied by hyporexia and night sweats. During the examination in the endocrinology office, a mass was palpated in the right iliac fossa, so the patient was referred to the emergency department. Ultrasonographic study identifies a rounded, heterogeneous, predominantly hypoechoic image, with a rounded calcification of regular and defined edges, located in the ileocecal valve in close relationship with iliac vessels. It measures 4.5 × 6.8 × 8.1 cms in its main axes; in the color Doppler examination it shows central and peripheral vascularization (Figure 1). Subsequently, CT of the abdomen with intravenous contrast medium showed an inflammatory process in the intestinal loops secondary to subacute-chronic infection located in the ileocecal region (Figure 2). In the evaluation by pediatric oncology, tumor markers and biopsy were ordered, but the procedure was deferred due to a positive PCR result for SARS-CoV-2.

[1]Resident Doctor of Specialization in Diagnostic and Therapeutic Imaging. Department of Diagnostic and Therapeutic Imaging, Hospital Centro Médico Nacional 20 de Noviembre, Mexico City, Mexico.

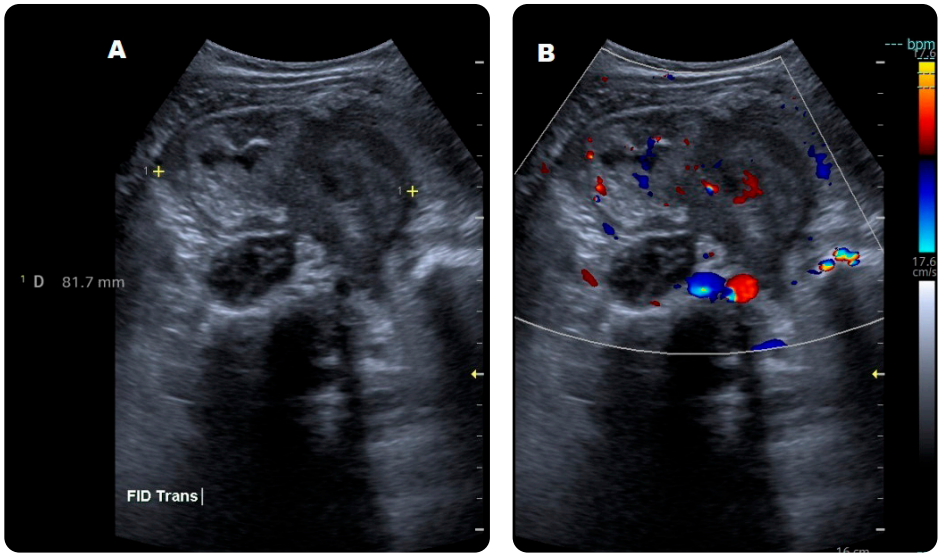


Figure 1. a) Gray scale ultrasound and b) application of color Doppler function, in axial section of the right iliac fossa. A rounded heterogeneous image of hypoechoic predominance with regular and defined borders is observed in the ileocecal valve, in close relation to iliac vessels; in the color Doppler scan it shows central and peripheral vascularization.

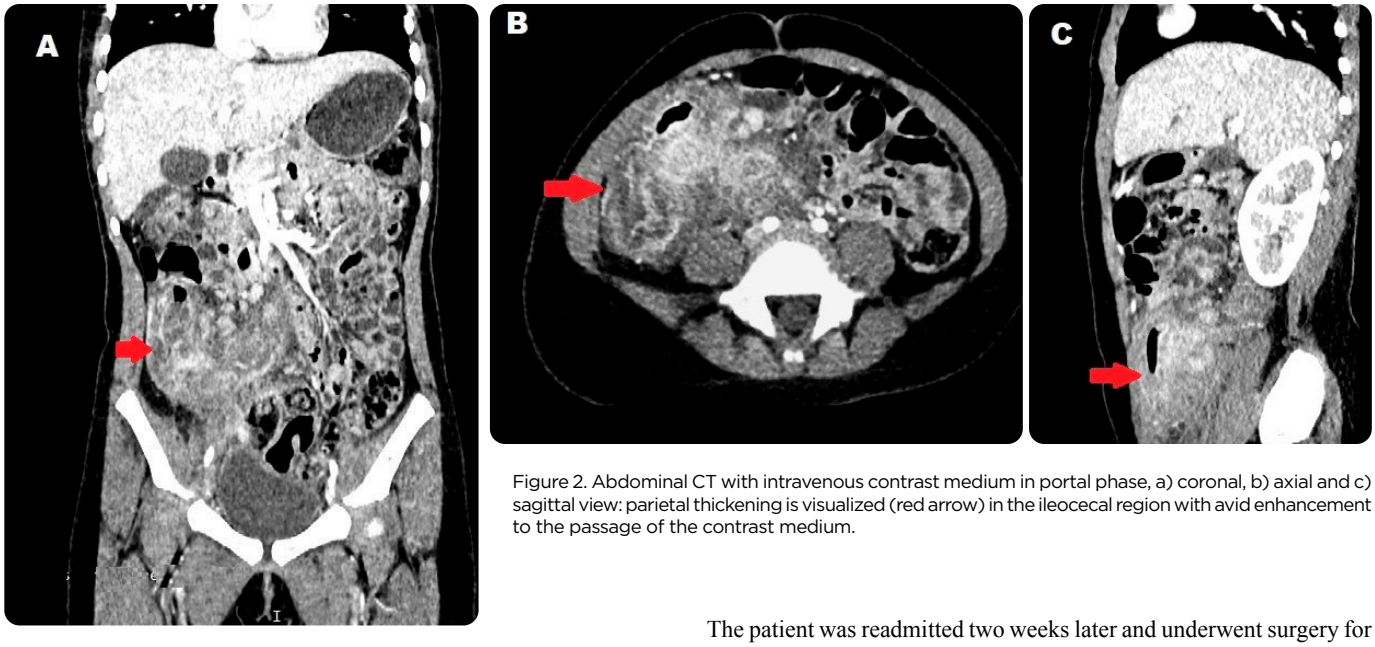


Figure 2. Abdominal CT with intravenous contrast medium in portal phase, a) coronal, b) axial and c) sagittal view: parietal thickening is visualized (red arrow) in the ileocecal region with avid enhancement to the passage of the contrast medium.

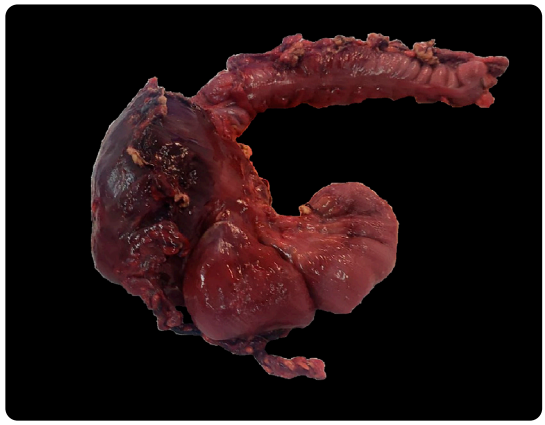


Figure 3. Right hemicolectomy resection specimen: ischemic and hemorrhagic areas are visualized in cecum and ileum, with mass appearance in cecal topography.

The patient was readmitted two weeks later and underwent surgery for removal of the intestinal tumor in the cecum plus resection of 20 cms of distal ileum, en bloc right hemicolectomy, termino-terminal ileocolic anastomosis, and sending of samples to pathology (Figure 3). Pathology reports were chronic active perforated enteritis with reparative plastron, fibrosis and granulation tissue associated with Actinomyces spp. No evidence of malignancy was observed.

Discussion

Actinomycosis is a chronic suppurative disease caused by Actinomyces species, which are members of the endogenous flora, normally found in the mouth, gastrointestinal tract and female genital tract. The clinical and radiological findings of actinomycosis are variable, depending on the primary site of involvement and the duration of the disease. In fact, it can often resemble other inflammatory and neoplastic lesions, so actinomycosis poses a diagnostic challenge,

especially when the clinical symptoms are not typical. For this reason, understanding the various imaging findings of actinomycosis grouped by sites of involvement is necessary for early diagnosis and appropriate initial antibiotic therapy to prevent extensive local tissue destruction and minimize surgery.

On average, about 20% of patients with actinomycosis present with the abdominopelvic form of the disease. Abdominal actinomycosis may go unnoticed for months or years due to its painless nature and nonspecific symptoms. Actinomycotic infection preferentially affects the ileocecal region of the bowel and spreads to surrounding structures through fascial and connective tissue planes. The aggressive nature of the infiltration is the telltale key to actinomycosis.

In cases of actinomycotic disease involving the bowel, the common CT finding is concentric wall thickening accompanied by a cystic or solid mass adjacent to the affected bowel segment. Such a mass is characteristically surrounded by prominent inflammatory infiltrates, and because of the extensive and dense fibrosis (hallmark of actinomycosis), the solid portions of the mass and surrounding soft tissue fibers around it tend to show marked enhancement after contrast administration. The invasive nature of the mass often suggests a tumor; however, regional lymphadenopathy is rarely associated with abdominal actinomycosis. Interestingly, despite its unlimited infiltrative nature, it is unlikely to spread to the peritoneal cavity. Ascites is usually minimal or may be absent (3).

CT is the diagnostic modality of choice to assess the degree of involvement and to verify the effectiveness of treatment, especially when this is not surgical. This disease tends to manifest as a solid mass with low focal attenuation, frequently with thin-walled cystic areas and produces infiltration.

The definitive diagnosis of actinomycosis requires histopathological study, in which sulfur granules and Gram-positive branching hyphae are observed.

Associated with the histopathological study, the standard diagnostic method is microbiological culture. However, this has certain limitations, since it is required to process the fresh material immediately or transport it in anaerobic containers; and even if treated with an adequate technique, it can give negative results in up to 76% of the cases (4).

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Correspondence

Juan P. Manrique-Salas
Avenida Félix Cuevas 540, Col del Valle Sur, Benito Juárez
Código postal 03229
Ciudad de México, México
jpmansa@gmail.com

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