

# Colonic pseudotumor caused by abdominal actinomycosis: A case report

## *Pseudotumor colónico causado por actinomicosis abdominal: reporte de un caso*

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### Abstract

We report a case of abdominal actinomycosis, a chronic suppurative infection caused by bacteria of the genus *Actinomyces*, simulating colon cancer, and presenting with abdominal pain and leukocytosis. Computed tomography revealed a mass lesion with irregular contours, infiltrative aspect, with extension to omental fat and abdominal wall, in the transverse colon. A surgical intervention was performed due to the suspicious of a colonic tumor. In the post-operative period, anatomopathological examination showed suppurative nodules associated with actinomycetes colonies, confirming the diagnosis of abdominal actinomycosis. After surgery, the patient was submitted to antibiotic treatment and no relapse was observed.

**Key words:** Actinomycosis. Colonic neoplasms. Abdominal abscess. Surgery

### Resumen

Reportamos un caso de actinomicosis abdominal, una infección supurativa crónica causada por bacterias del género *Actinomyces*, que simula el cáncer de colon y se manifiesta con dolor abdominal y leucocitosis. La tomografía computada reveló una lesión sólida con contornos irregulares y aspecto infiltrativo en el colon transverso, con extensión al epiplón y la pared abdominal. La intervención quirúrgica fue realizada debido a la sospecha de un tumor de colon. En el posoperatorio, el examen anatomopatológico mostró la presencia de nódulos supurativos asociados con colonias de actinomicetos, lo que confirma el diagnóstico de actinomicosis abdominal. Posteriormente a la cirugía el paciente recibió antibioticoterapia y no presentó Recidivas.

**Palabras clave:** Actinomicosis. Neoplasias de colon. Absceso abdominal. Cirugía.

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## Introduction

Gram-positive, facultative, or obligate anaerobic bacteria of the genus *Actinomyces* are the microorganisms responsible for the development of actinomycosis<sup>1,2</sup>. They are found only in body temperature in humans and animals. *Actinomyces* species are also prevalent in the soil, but these have not been identified as causing diseases in humans<sup>3</sup>. The species responsible for the disease in humans is, in most cases, *Actinomyces israelii*<sup>3,4</sup>, which is an endogenous inhabitant of most mucous membranes<sup>4,5</sup>. There is no person-to-person or animal-to-person transmission of this agent<sup>5</sup>.

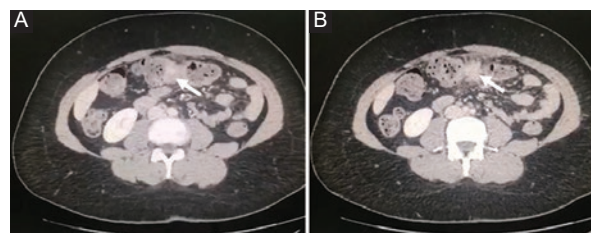
Actinomycosis is a chronic, indolent, and slow-progression infection<sup>2,6-8</sup>. This infection occurs in immunocompetent patients, having as a gateway the discontinuity of anatomical barriers, which allows the access of the commensal flora of the mucous membranes to the deep tissues, by contiguity. It leads to the formation of single or multiple abscesses that result in drainage sinuses<sup>2,4,6,7</sup> and are surrounded by granulation tissue and fibrosis<sup>4,5,9,10</sup>. The bacteria, when causing tissue injury, initiate a poorly irrigated granulation process, favoring their anaerobic development<sup>1</sup>. This lesion is called an inflammatory pseudotumor, which can mimic, clinically and radiologically, a malignant neoplasm<sup>1,4,10</sup>. Today, it is considered a rare infection, but it has been common in the pre-antibiotic era<sup>11</sup>.

The infection mainly affects the orofacial and cervicofacial regions since the bacteria are usually found in the oral cavity<sup>1,2,9</sup>. In these cases, it is usually associated with poor oral hygiene<sup>3</sup>. In addition, they are also found naturally in the gastrointestinal tract, bronchi, and female genital system. Abdominal involvement is rare, as is the involvement of the pelvic region, liver, bones, and perianal region<sup>8</sup>.

Because abdominal actinomycosis is less frequent and difficult to diagnose, and due to the complex differential diagnosis, the treatment is often late or iatrogenic<sup>1</sup>. The diagnosis is often posterior to a laparotomy followed by resection of the intestinal segment and anatomopathological study of the surgical specimen<sup>1</sup>. The present report is, therefore, extremely important to warn about the diagnostic possibility of abdominal actinomycosis.

## Case presentation

A 46-year-old female patient from Belo Horizonte/MG sought treatment with non-specific abdominal



**Figure 1. (A and B):** Computed tomography showing, by the arrows, the lesion suggestive of transverse colon tumor.

pain, with no apparent cause, worsening in the last 48 h. She also had sporadic dysuria. She denied fever, changes in intestinal habit, and weight loss. Arterial hypertension patient, with a 25-pack-year smoking history. Negative family history for neoplasms.

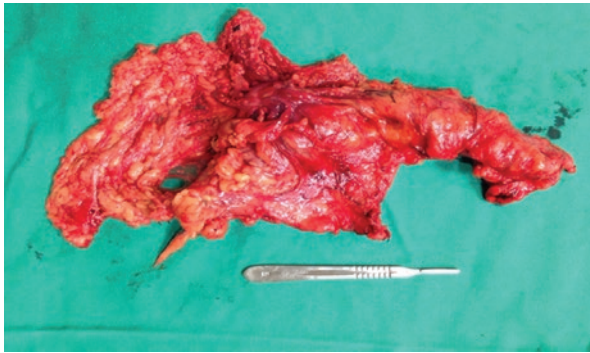
Laboratory tests showed leukocytosis ( $15.300/\text{mm}^3$ ) without left deviation and pyuria in the urine test. The hypothesis of urinary tract infection was suggested, and antibiotic therapy was performed with nitrofurantoin 300 mg/day (100 mg every 8 h) for 5 days.

After 1 week of treatment, abdominal pain worsened. Physical examination showed no abnormalities except for pain at deep palpation in the upper abdomen, but no signs of peritoneal irritation. At this time, a new blood count revealed increased leukocytosis.

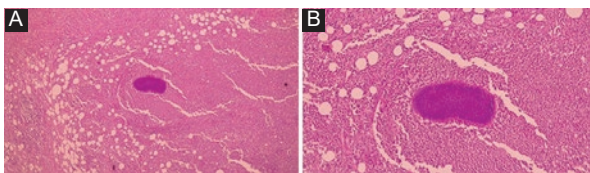
Computed tomography (CT) scanning of the abdomen (Fig. 1) revealed a contrast-enhancing mass lesion measuring  $3.2 \times 3.6 \times 2.8$  cm with irregular and poorly defined contours in the middle third of the transverse colon. It had an infiltrative aspect and extension to omental fat and anterior abdominal wall.

The patient, with diagnostic suspicion of gastrointestinal stromal tumor or adenocarcinoma of the colon, underwent diagnostic laparoscopy, which revealed a massive tumor in the transverse colon and omental invasion. Transverse colectomy followed by latero-lateral and colon-colonic anastomosis was performed. In addition, omentectomy and retroperitoneal lymphadenectomy were done to evaluate metastasis and cancer staging (Fig. 2). The patient had good post-operative evolution and was discharged after 2 days, being referred to the clinical oncology service.

The anatomopathological examination (Fig. 3) showed colonic pseudotumor lesion consisting of suppurative nodules associated with actinomycetes colonies with intact muscle lining. Epiploons and lymph nodes were negative for tumor infiltration and metastasis. Due to the result, the patient was again admitted to the hospital for infectology care. It was suggested intramuscular benzathine benzylpenicillin 12 million UI/day for 6 weeks



**Figure 2.** Anatomical surgical specimen showing the colonic pseudotumor.



**Figure 3. (A and B):** Hematoxylin-eosin stained light microscopy demonstrates abscess/inflammatory cells permeating the focus of actinomycosis. (B): with magnification.

followed by oral ampicillin 2 g/day for 3 months. After the whole treatment and 1-year follow-up, the patient had no evidence of relapse.

## Discussion

The general incidence of actinomycosis is virtually impossible to ascertain since failure to consider it as a diagnosis and difficulties in proving it lead to under-reporting<sup>3</sup>. No recent estimates of the disease occurrence have been found, but historically, 20-40 cases are reported in the United Kingdom each year<sup>3</sup>.

Abdominal actinomycosis represents about 20% of all cases of actinomycosis<sup>1,2,6</sup>. The disease classically presents as a slow-growing tumor, usually in the ileocecal region (65%), and less frequently in the stomach, duodenum, liver, and rectum<sup>4</sup>. Abdominal actinomycosis is approximately 3 times more common in men than in women, although an increasing number of cases occur in association with the use of intrauterine contraceptive devices<sup>3,11</sup>. There is no correlation between the disease and place of residence, social class, or ethnicity<sup>3,9</sup>. The condition is predominant in the fourth and fifth decades of life; the infection is rare in children and in patients over 60 years of age<sup>1,2</sup>. In HIV-positive or immunosuppressed patients, more invasive and necrotizing forms develop<sup>1</sup>.

The mechanism of pathological infection is unclear, but circumstantial evidence suggests that prior mucosal injury is necessary before actinomycosis occurs. Perforated appendicitis is the most common cause, followed by neoplasia or trauma, as well as perforation of the gastrointestinal tract by a foreign body<sup>3</sup>. Other factors predisposing to its development include previous abdominal surgeries, colonic diverticulitis, mesenteric vascular insufficiency, and cesarean sections<sup>2,4,6,7</sup>. Occasional cases were reported without prior mucosal injury<sup>3</sup>.

Actinomycotic infection is characterized by a progressive inflammatory response that is contiguous and insidious, with multiple connected abscesses and an aggressive desmoplastic process<sup>3</sup>. Abscesses usually consist of a thick layer of granulation tissue around a central pus reservoir. The granulation zone is highly cellular, including fibroblasts and collagen fibers, and the central pus zone contains the typical sulfur granules with occasional liquefaction<sup>3</sup>. Diagnostic suspicion is due to the finding of these sulfur granules in the anatomopathological examination. Sulfur granules represent *Actinomyces* colonies, and, although highly suggestive, they are not pathognomonic of actinomycosis<sup>1,2,6,9</sup> since similar observations can be found in infections by *Nocardia brasiliensis*, *Streptomyces madurae*, and *Aspergillus*<sup>4,10</sup>.

The infection spreads by proximity and infiltrates adjacent tissues and organs. The lesions are very infiltrative due to the secretion of proteolytic enzymes that allow them to cross anatomical barriers<sup>12</sup>. Spread is rarely lymphatic or hematogenic<sup>3,9</sup>. For the most part, actinomycosis presents as a mixed infection, in which other bacteria other than *A. israelii* can be isolated. It has been suggested that *Actinomyces* are pathogenic only with the synergistic action of other bacteria<sup>3,5</sup>.

Pre-operative clinical and radiological diagnosis is rarely performed<sup>9</sup>. The clinical picture of abdominal involvement presents non-specific signs such as fever, weight loss, constipation or diarrhea, nausea, vomiting, anorexia, and abdominal pain<sup>1,2,6</sup>, besides the presence of abdominal mass and anemia<sup>1</sup>. Occasionally, leukocytosis is found<sup>9</sup>, as in the case reported. In radiological terms, CT reveals an infiltrating and non-homogeneous mass. The masses due to actinomycosis appear more frequently as predominantly solid lesions or as mixed cystic and solid lesions, with blurred margins and irregularly thickened walls<sup>12</sup>. In addition, it allows the definition with greater precision of the dimensions and extent of the

infection<sup>6</sup>. CT can also be used as a guide for percutaneous biopsy and abscess drainage, which would aid in a pre-operative diagnosis<sup>1,2,6</sup>. However, the diagnosis of actinomycosis is rarely considered. Most cases of abdominal actinomycosis are discovered randomly during surgery<sup>9</sup>.

If the diagnosis is made early and the antibiotic therapy is adequate, the prognosis is good<sup>1</sup>. Despite this, some case reports described in the literature demonstrate higher rates of disease recurrence when antibiotic treatment is not accompanied by surgical resection of the affected areas<sup>9</sup>. In case, the finding was made during surgery, even if no microbiological culture is made, if the anatomopathological examination shows the sulfur granules, the antibiotic treatment should be initiated<sup>9</sup>.

*Actinomyces* are sensitive to most antibiotics but are less sensitive to metronidazole<sup>9</sup>. The treatment of choice is penicillin-based therapy. Tetracycline, erythromycin, or clindamycin can be used in patients allergic to penicillin<sup>1,2,6,7</sup>. Intravenous penicillin G at doses of 20 million IU/day for 4 weeks is the drug of choice, followed by oral penicillin V at doses of 2-4 g/day for 2-12 months<sup>9</sup>. Surgical treatment associated with antibiotic therapy shows a cure rate > 90%<sup>1</sup>. The extent of surgical resection must follow the criteria of oncologic surgeries<sup>9</sup>.

Despite the good prognosis, it is possible to notice that abdominal actinomycosis remains a challenge in terms of clinical diagnosis. It is an uncommon condition that mimics a wide variety of abdominal complaints of acute and inflammatory characteristics. Although actinomycosis is a rare entity, it should always be included in the differential diagnosis of abdominal disease with tumors of inflammatory characteristics that tend to invade adjacent tissues and structures. Obtaining an accurate diagnosis is essential for the successful management of abdominal actinomycosis.

## Conflicts of interests

The authors declare that there have no conflicts of interest in this work.

## Ethical disclosures

**Protection of human and animal subjects.** The authors declare that no experiments were performed on humans or animals for this study.

**Confidentiality of data.** The authors declare that they have followed the protocols of their work center on the publication of patient data.

**Right to privacy and informed consent.** The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

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