SURGERY

Two case reports of mesenteric and retroperitoneal actinomycosis and a narrative review of the relevant literature

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Abstract

Introduction. Actinomycosis is an uncommon subacute or chronic suppurative bacterial granulomatous infectious disease with clinical heterogeneity. The majority of actinomycosis cases were of extra-abdominal origin, with oro-cervico-facial cases representing 55%, abdominopelvic representing 20%, and thoracic representing 15% of total reports. Currently, abdominal actinomycosis incidence is approximately 1 case per 119,000 people, being found three times more frequently among males. We report two rare clinical presentations of abdominal actinomycosis affecting the mesentery and the retroperitoneum, respectively.

Case Report 1. A 58-year-old Caucasian male presented to our clinic with abdominal pain in the right upper quadrant. Pre-operative evaluation, although inconclusive, showed a mesocolic mass infiltrating the right and transverse colon. The patient underwent exploratory laparotomy. After partial resection of the mass, the histopathology report demonstrated mesenteric actinomycosis.

Case Report 2. A 40-year-old Caucasian male presented to our clinic complaining about a mucopurulent material from an orifice at the right inguinal region. After appropriate work-up, a large abdominopelvic, stellate mass (75 x 22.8 mm) in the retroperitoneum was revealed. Surgery along with the appropriate antibiotics was used to treat the patient.

Conclusion. Preoperative suspicion and diagnosis of actinomycosis are very challenging, with a high rate of misdiagnosis often resulting in delayed treatment. Our case reports highlight that abdominal actinomycosis should always be part of differential diagnosis, especially when there is involvement of multiple organs. The gold standard treatment of actinomycosis is surgical excision with prolonged antibiotic treatment.

Keywords: actinomyces, actinomycosis, retroperitoneal, mesenteric, abdominal, mesocolic mass

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Introduction

Actinomycosis is an uncommon subacute or chronic suppurative bacterial granulomatous infectious disease with clinical heterogeneity caused by Actinomyces species, a group of anaerobic or microaerophilic non-spore forming Gram-positive bacteria. The main pathogens of this species responsible for human actinomycosis are Actinomyces israelii and less commonly Actinomyces gerencseriae [1,2]. Actinomyces israelii normally inhabits the oral cavity, pharynx, skin, gastrointestinal and urogenital tract [3]. Depending on the body system affected, actinomycosis can be classified as oro-cervico-facial, thoracic or abdominopelvic disease. The involvement of other body regions

leads to symptoms from the central nervous system (CNS), the cardiovascular system, the musculoskeletal system, the urinary system, and the skin [3,4]. Lately, a decreasing incidence of the oro-cervico-facial type of actinomycosis has been reported, whereas abdominal and pelvic infections are increasing, mainly due to the use of intrauterine contraceptive devices (IUCD) [5].

We report two rare, clinical presentations of abdominal actinomycosis affecting the mesentery and the retroperitoneum, respectively. The following case reports are presented in accordance to Surgical CAse REport (SCARE) guidelines [6]. We also review the relevant literature regarding the presentation and overall management of abdominal actinomycosis.

Case reports

Case 1

A 58 -year-old Caucasian male presented with a 2-week history of intense abdominal pain at the right upper quadrant, no nausea or vomiting, no alteration of defecation, and only a few days' history of decreased appetite and food intolerance. The blood test results revealed elevated inflammatory markers (65 mm ESR, 0.2 mg/dl procalcitonin, and 5.24 mg/dl CRP), white blood cell count (13.34 k/μl with 71.0% neutrophils, 11.1% lymphocytes) and normocytic anemia with 9.5 g/dl hemoglobin, 28.2 hematocrit, and 87 fl MCV.

Computed tomography of the abdomen showed a tumor-like, infiltrative mass at the right colon that obstructed the lumen and expanded to the surrounding mesentery. It also revealed enlarged lymph nodes at the right colon mesentery, a thickening of the anterior pararenal peritoneum along with a few suspicious lymph nodes in the retroperitoneum.

Colonoscopy ruled out an intraluminal mass, but obstructing edema at the right colon was observed and biopsies were taken (Figure 1a, 1b). Histology report showed inflammatory infiltrations of leukocytes, plasmacytes, and eosinophils. Gastroscopy was also performed to rule out a duodenal origin of the mass. Due to the non-conclusive findings, an MRI investigation of the abdomen was performed, reporting a right colon mass with features suggestive of a neoplasm (Figure 2a, 2b).

Taking the above findings into consideration, the decision was made for an exploratory laparotomy. The intraoperative findings were a mesocolic mass infiltrating the right and transverse colon, pressing the duodenum. Another mass was also palpated in the retroperitoneal area, albeit out of our visual field. Fast track biopsy revealed an inflammatory mass with the presence of actinomycetes. A decision was made to continue with partial resection of the mass along with the right colon.

The final histopathologic report showed an external infiltrative mass that extended to the serosa and muscle layer of the colon. Actinomyces were also inspected within the surrounding lymph nodes (Figure 3a, 3b). Intraoperative microbiology culture was negative for anaerobes. The patient was administered a targeted antibiotic therapy with high dose IV penicillin G. His postoperative course was uneventful and was let to leave the hospital after 8 days, at his request, but against medical advice.

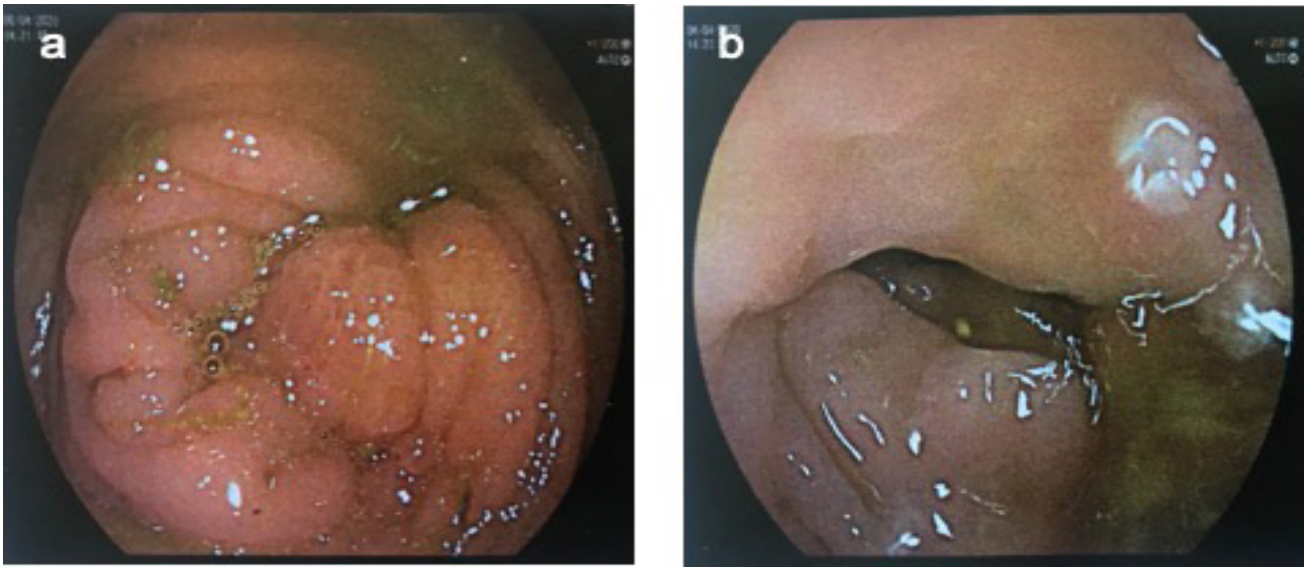

Figure 1a, 1b. Colonoscopy ruled out an intraluminal mass, but obstructing edema at the right colon was observed.

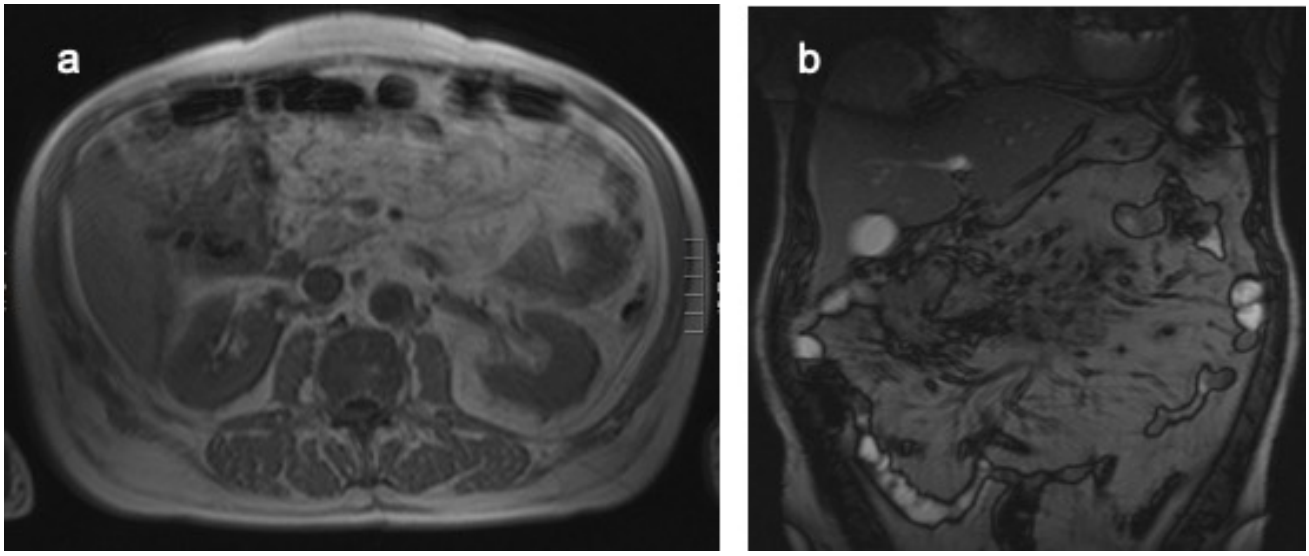

Figure 2a, 2b. An MRI scan showed an infiltrative mass at the right colon that obstructed the lumen and expanded to the surrounding mesentery with enlarged lymph nodes at the right colon mesentery.

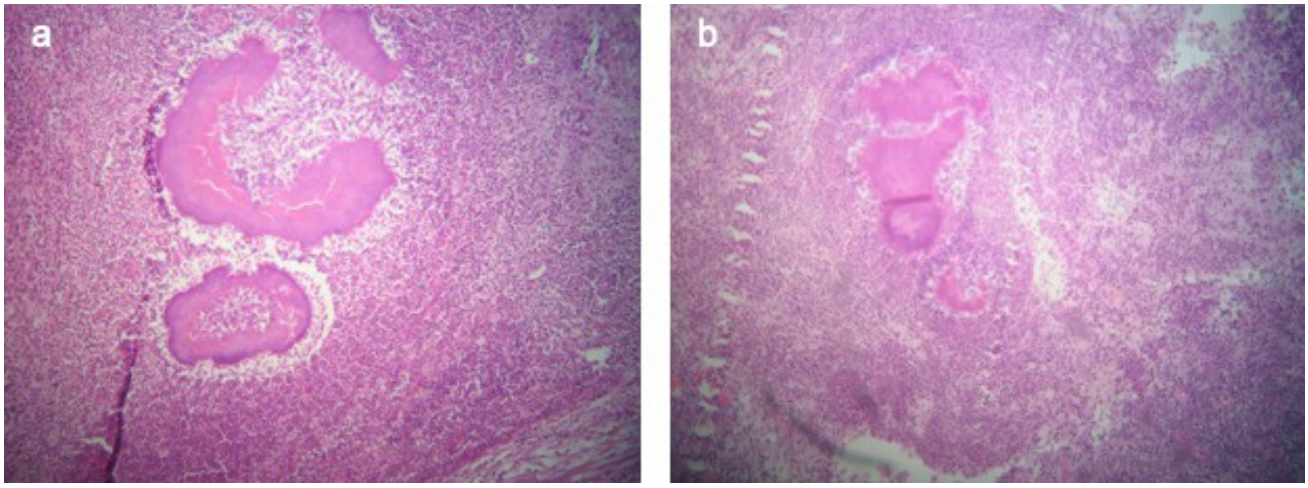

Figure 3a, 3b. Histopathologic report showed an external infiltrative mass that extended to the serosa and muscle layer of the colon. Actinomyces were also inspected within the surrounding lymph nodes.

Case 2

A 40-year-old white male, weighing 73 kg, presented to our clinic complaining about a mucopurulent material from an orifice at the right inguinal region. The patient did not experience any fever, night sweats, weight loss, or movement restriction. Past medical history was negative for any illnesses. Of note, the patient had a crush accident with multiple injuries eight years ago and a pancreatic pseudocyst, which was drained ten years ago, percutaneously. On physical examination, the abdomen was soft, not tender, and vital signs were within normal range. Liver and renal function assays were normal. Urine analysis and chest x-ray were normal. White blood count was 7.9 k/μl with 63.3% neutrophils, 5.0% lymphocytes, 27.0% monocytes, 2.1% eosinophils and 2.6% basophils, hematocrit was 41.1%. C-reactive protein and erythrocyte sedimentation rate were normal. The patient was given a second-generation cephalosporin (cefaclor) and was discharged home while awaiting further investigation.

During further work-up, an abdominal sonogram showed one hypoechoic, space-occupying lesion in the right retroperitoneal space. A retroperitoneal neoplasm was suspected, and a computed tomography (CT) scan was obtained with intravenous and oral contrast-enhanced material.

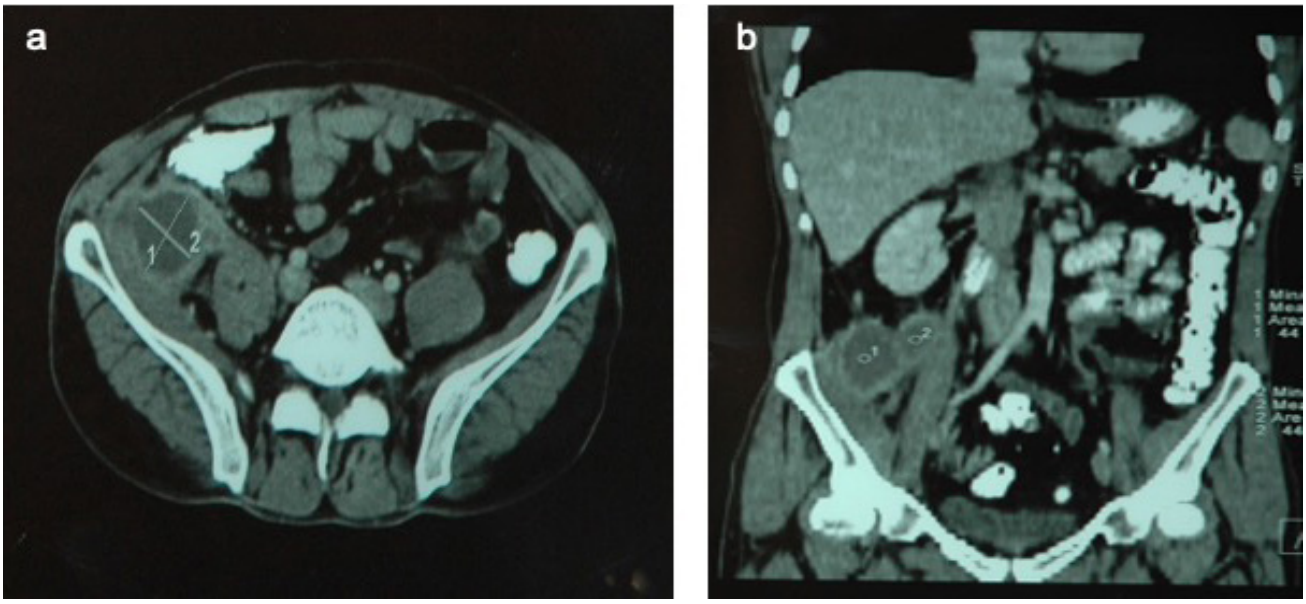

Figure 4a, 4b. Abdominopelvic, stellate mass (75 x 22.8 mm) in front of the psoas muscle, extending from the lower pole of the right kidney to the pelvis and spreading superficially in the right inguinal region, ultimately forming a fistula.

CT scan evidenced a large abdominopelvic stellate mass (75 x 22.8 mm) in front of the psoas muscle which was extending from the lower pole of the right kidney to the pelvis and was spreading superficially in the right inguinal region forming a fistula (Figure 4a, 4b). No other pathologies from the abdominal organs were identified.

The patient was referred to the operation room for exploratory laparotomy. The mass was removed, and tissue samples were extracted. General anesthesia was utilized without any perioperative adverse event. Histological examination revealed no evidence of malignancy. Chronic inflammation with granulation tissue and foamy macrophages was reported. Further examination revealed sulfur granules, consistent with actinomycosis. Intravenous penicillin treatment was administered for two weeks, and the patient was discharged home with oral ampicillin with sulbactam for six months.

A month after the surgery, a fistulography showed closure of the canal with minimal discharge. Postoperative three-month follow-up with a series of echo and CT scans demonstrated an improvement of the findings with progressive decrease in the size of the retroperitoneal abscess, until the resolution of the mass at the six-month follow-up.

Discussion

Actinomyces spp. are Gram-positive filamentous anaerobic or microaerophilic rod-shaped bacteria and do not belong to the group of fungi [7]. The majority of actinomycosis cases were of extra-abdominal origin, with oro-cervico-facial cases representing 55%, abdomino-pelvic representing 20%, and thoracic representing 15% of total reports [8,9]. Currently, abdominal actinomycosis incidence is approximately 1 case per 119,000 people, presenting three times more frequently among males.

The first report of abdominal actinomycosis goes back in 1846 by Bradshaw. Actinomyces israelio is usually part of the normal flora of the oral cavity, bronchi, gastrointestinal and female genital tract [10]. It has low virulence potential and requires a disruption of the mucosal barriers or/with decrease in host's immune system capacity [11] to invade surrounding tissues. In general, abdominal actinomycosis is common among patients with poor general health and comorbidities [12]. Predisposing factors include acute appendicitis with perforation (main predisposing factor [10]), localized inflammation, abdominal trauma, peptic ulcer disease [13], prolonged intrauterine contraceptive device (IUCD) maintenance, abdominal surgical or endoscopic interventions [14], foreign bodies [15], peritoneal dialysis, neoplasms, poor oral hygiene [9], use of T-tube during abdominal surgeries in combination with factors inducing immunosuppression [16]. Our second patient's medical history with retroperitoneal actinomycosis presented with many predisposing factors, such as multiple injuries from trauma and pancreatic pseudocyst drainage. Since actinomyces is a slow-growing bacterium, intrabdominal manifestations can be presented even many years after mucosal disruption [8].

Intra-abdominal structures mostly affected are

the ileocecal area, the appendix, and the right colon, whereas the left colon is rarely affected [17]. Abdominal actinomycosis is characterized by infiltrative and granulomatous inflammation, with the formation of granulation tissue, abscesses, and sinuses, pus production, followed by necrosis and reactive fibrosis, while the central area of pus reveals the typical sulfur granules on histological examination [10]. Nevertheless, sulfur granules are found only in 50% of actinomycosis cases and are not pathognomonic [18]. Fistulization has been reported during the late stages of inflammation, both internally and towards the skin [5,10]. The drainage of mucopurulent material from an orifice at the right inguinal region that occurred in our patient pointed toward the diagnosis of retroperitoneal actinomycosis. A great variety of other microorganisms are also detected in cultures, depending on the site of infection, thus enhancing the perception of synergy as a mechanism of actinomycosis infection [10]. The macroscopic image may mimic inflammatory bowel disease, acute appendicitis, carcinoid, malignancies, tuberculosis, pelvic inflammatory disease, tubo-ovarian abscess, and amoebadosis [14,17,19-22]. Hepatic actinomycosis comprises up to 5%-15% of abdominal case reports and usually manifests as multiple abscesses [17]. Renal, gallbladder and biliary tract [23], pancreas, stomach, small intestine, and omental actinomycosis cases have also been reported [24–26].

Mesenteric actinomycosis is extremely rare, with very few reports in the literature, and usually presents as a mesenchymal mesenteric tumor with non-specific characteristics [4]. Chan et al. described the case of a 45-year-old man with abdominal pain, whose CT image revealed a mesentery-limited actinomycotic abscess without extension to surrounding tissue, except for the accompanying inflammation of the appendiceal tip [20]. Similarly, Segovia-García et al describe the case of a 55-year-old female complaining about abdominal pain and a palpable mass. Diagnostic approach led to the preoperative diagnosis of neoplasia, while histological examination revealed mesenteric actinomycosis, highlighting the insufficiency of radiological and laboratory tests in diagnosing this pathologic entity [11].

Retroperitoneal actinomycosis also needs to be a suspicion in case of retroperitoneal masses and the presence of predisposing factors since this entity is usually masqueraded by the diagnosis of neoplasia. Berchtenbreiter et al. presented the case of a young patient with actinomycosis, whose primary diagnosis led toward adrenal pheochromocytoma [12]. We should highlight that the most common finding in the retroperitoneum is the presence of firm, fibrous, avascular tissue, which can lead to pitfalls during laparoscopy [12]. In our case, a large abdominopelvic, stellate mass with the characteristics in front of the psoas muscle was found intraoperatively. Retroperitoneal infection sites can be attributed to hematogenous or direct spread [17].

Preoperative accurate diagnosis of actinomycosis is reported in less than 10% of patients infected, due to the lack of clinical suspicion and non-specific symptomatology [19]. Initial symptoms are usually non-specific [16,17,19]. More specific clinical signs and symptoms depend on the region affected. The presence of an intra-abdominal solid mass and the absence of leukocytosis or elevation of inflammation markers can lead to misdiagnosis of malignancy preoperatively in some patients. There are no specific radiological or endoscopic features indicating adnominal actinomycosis and special attention should be paid to findings of infiltration and formation of fibrotic tissue, abscesses, colonic wall thickening, and mass formation on CT [10]. CT-guided aspiration and core biopsy could contribute to diagnosis preoperatively if sulfur granules are present and cultures are positive, when actinomycosis is suspected [19]. The utility of MRI in actinomycosis investigation has not been established yet, but findings may reflect the stages of actinomycosis infection [5]. Preoperative colonoscopy is recommended in many patients, to exclude colitis or malignancy. Corresponding to the patient of the first case presented here, preoperative colonoscopy ruled out an intraluminal mass and revealed obstructing edema at the right colon. Diagnosis is aided by histopathological examination, which reveals characteristic actinomycotic mycelium-like shape sulfur granules, while the Gram-positive stain presents multiple branching swirling Gram-positive rod-shaped bacteria [5]. Microbiological culture remains the most useful diagnostic tool, giving characteristic colonies with a hard molar tooth-like appearance. Moreover, culture confirmation of actinomycosis is rare among literature case reports, such as in our case reports, since actinomycosis was not included in the differential diagnosis at the time of surgery [5].

If actinomycosis is diagnosed by preoperative test or by intra-operative frozen section and the infection course is uncomplicated, high-dose antibiotic therapy for a prolonged period, due to poor drug penetration through fibrotic tissue, is the treatment of choice. Limited en-bloc surgical excision of the mass, combined with antibiotic agents may pose as a definitive treatment [10,17]. In the preoperative or intraoperative confirmation of actinomycosis, there is no need to apply oncological resection principles. If there is no culture confirmation, but the histological examination reveals sulfur granules, antibiotic treatment should start immediately [5]. Additionally, the administration of metronidazole against Gram-negative anaerobic normal flora, which is thought to act synergistically in actinomycosis infection, is also reported [5]. CT findings may be useful both for detecting residual mass or recurrence, but also for deciding the

duration of therapy based on individual response [19]. Mortality rates are extremely low and prevention of recurrence is based on long-term antibiotic schema in combination with initial surgical treatment, which is effective in more than 90% of the cases with good prognosis [5].

Conclusions

Preoperative suspicion and diagnosis of actinomycosis are very challenging, with a high rate of misdiagnosis towards malignancy, often resulting in delayed treatment. It is worth mentioning that this entity is frequently referred to as the great imitator in literature. Our case reports highlight that abdominal actinomycosis should always be part of differential diagnosis, especially when infiltration of multiple compartments and organs is present. When actinomycosis is confirmed, surgical excision of intrabdominal lesion combined with prolonged antibiotic treatment is of great importance.

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