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journal homepage: www.casereports.comPara-iliac actinomycetoma presenting as sarcoma, a late complication of appendicitis: A case report[☆]Pierre Navarre^{a,b,c,1}, Marie-Andrée Cantin^{a,c,*}, Marc H. Isler^{a,b,c,2}^a Hôpital Sainte-Justine, Montréal, QC, Canada^b Hôpital Maisonneuve-Rosemont, Montréal, QC, Canada^c Université de Montréal, Montréal, QC, Canada

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ABSTRACT

INTRODUCTION: Actinomycosis is known to mimic several types of neoplasms, leading to morbid surgical interventions.**PRESENTATION OF CASE:** We report the particular case of an extensive right para-iliac actinomycetoma presenting as a sarcoma, which to our knowledge has not yet been described in the literature, in a patient with previous ruptured appendicitis. Thanks to the collaborative work between the orthopedic and general surgeons, pathologist and microbiologist, the diagnosis of actinomycosis was made pre-operatively, saving this 15-year-old patient from a tumor resection protocol.**DISCUSSION:** Actinomycetomas have often been reported to present in the same way as several abdominal and gynecological neoplasms, and on rare occasions been described as mimicking other soft-tissue sarcomas, leading to unnecessary morbid tumor resection protocols. The most common cause of abdominal actinomycosis is perforated appendicitis, and may present several years later.**CONCLUSION:** While faced with a soft tissue mass transgressing tissue planes and possibly extending to the region of the right lower quadrant, especially with a history of previous perforated appendicitis, one should consider the possibility of an abscess caused by pathogens of intestinal origin, including the gram positive anaerobe *Actinomycosis israelii*. Failure to actively search for this pathogen, which is not detectable with routine staining techniques and may take up to 1–2 weeks to isolate, may lead to unnecessary morbid surgical procedures.

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1. Introduction

Actinomycosis israelii is a gram positive anaerobic or microaerophilic bacterium of low virulence, and is part of the oral, bronchial, digestive and female genital flora. Whereas the infectious cases caused by this pathogen are rare and predominate in the maxillofacial area, abdominal infections are even rarer, often appearing following perforated appendicitis. As its presentation is variable, actinomycosis is well known to mimic several types of neoplasms, leading to morbid surgical interventions.

We present the case of a para-iliac actinomycetoma presenting as a sarcoma, which to our knowledge has not yet been described, in a patient with past history of appendicitis, followed by a review of the literature.

2. Presentation of case

We conducted a case study of a 15-year-old female patient presenting with a right para-iliac mass. This patient had no past medical history other than perforated appendicitis two years earlier, treated by open appendectomy and intravenous antibiotics for a week, without any subsequent symptoms. She described right flank pain of two months duration, with a progressively enlarging mass in the same region. Furthermore, she reported asthenia, a worsening limp and intermittent low-grade fever. Physical examination revealed a non-toxic patient with a large painful right iliac mass. There was no adenopathy.

Radiological investigation including plain X-rays, CT-scan and MRI revealed a soft-tissue mass with a necrotic center, extending on both sides of the right iliac wing, without any osseous involvement. This mass measured 7.5 cm × 3 cm internally and 8.5 cm × 4 cm externally with respect to the iliac wing (Figs. 1 and 2). Blood work revealed leucocytosis of 18.6 × 10⁹/L

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Fig. 1. (A and B) Axial CT scan images demonstrating the mass on both sides of the right iliac wing, without osseous involvement, with a necrotic center.

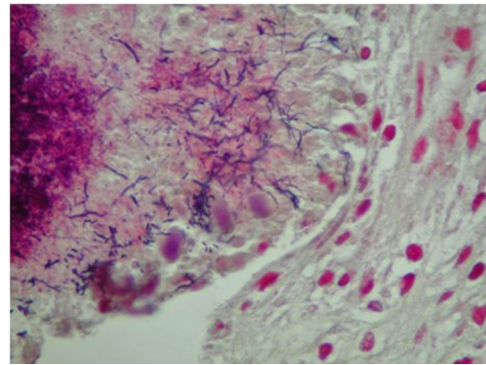


Fig. 3. Gram stain revealing the gram-positive bacilli *Actinomyces israelii*.

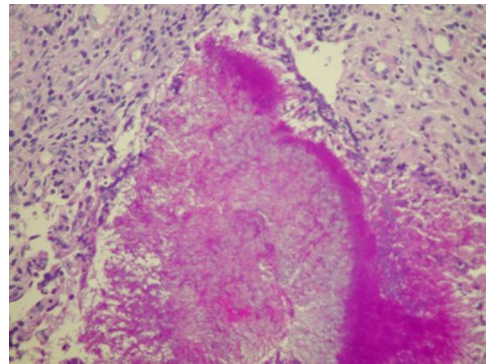


Fig. 4. "Sulfur granules" characteristic of actinomycosis.

and elevated inflammatory markers (erythrocyte sedimentation rate of 50 mm/h and C-reactive protein of 150 mg/L). Metastatic work-up did not reveal any abnormalities.

Faced with this clinical picture, our differential diagnosis included sarcoma, such as Ewing's sarcoma, and lymphoma. A bacterial abscess was considered less likely because of the slow progression and lack of systemic toxicity.

Core needle biopsy was performed and, thanks to a thorough search with special coloration techniques and collaborative work between microbiologist and pathologist, revealed gram-positive bacilli and "sulfur granules" suggestive of actinomycosis (Figs. 3–5). There was no evidence of neoplasia. Thus, we undertook surgical debridement and discovered an impressive multilocular abscess, extending from the ileo-caecal region to the gluteal region, and on either side of the iliac wing, sparing the iliac bone. The final cultures confirmed the diagnosis of actinomycosis, along with several intestinal co-pathogens. This patient was treated with oral antibiotics for a total of 8 months and required a second

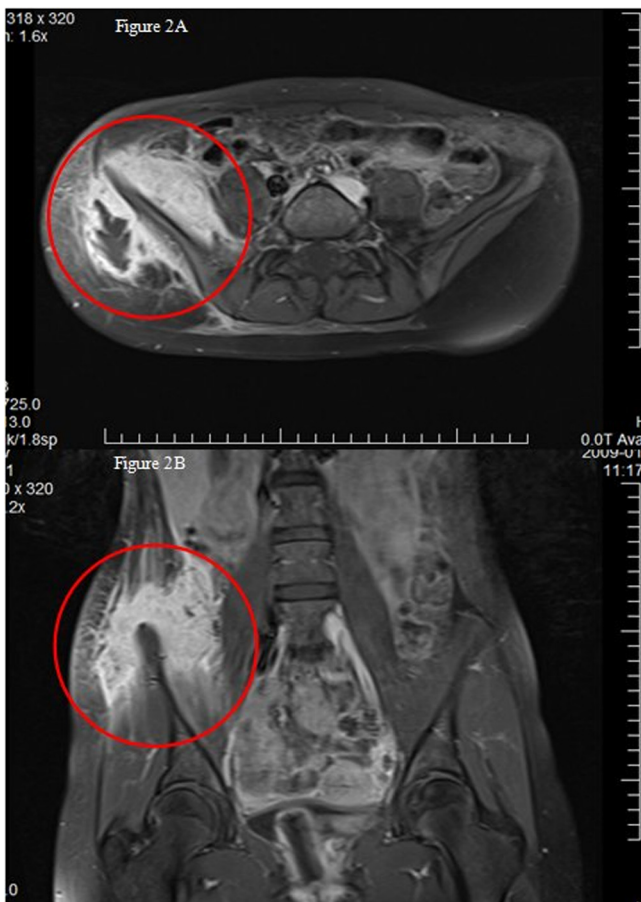


Fig. 2. (A and B) T1-FS MRI images (axial and coronal) demonstrating the mass on both sides of the right iliac wing.

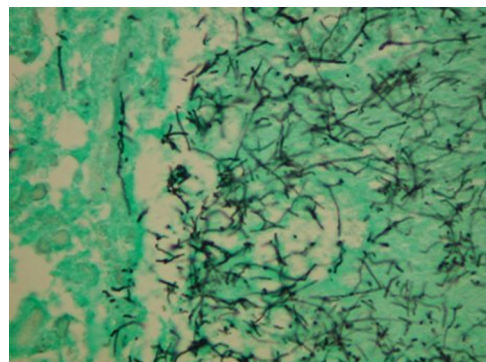


Fig. 5. Grocott argentation technique demonstrating the bacilli of actinomycosis.

surgical debridement 2 months after the first intervention. At one-year follow-up, she was doing very well, without any residual symptoms, and all investigations confirmed absence of disease.

Thanks to the collaborative work between the orthopedic and general surgeons, the microbiologist and the pathologist, a morbid surgical tumor resection protocol was avoided in this patient in the context of a clinical presentation suggesting a soft tissue sarcoma.

3. Discussion

A. israelii, the predominant form in human infection, is an anaerobic or microaerophilic gram-positive bacillus, which is part of the oral, bronchial, digestive and female genital flora. The infectious cases caused by this pathogen are rare and predominate in the maxillofacial area (more than 50%), however thoracic and abdominal infections are also seen (nearly 20% each).^{1,2} Several other anatomic locations have been reported sporadically.

Because of its low virulence, a mucus membrane breach, the presence of devitalized tissue and the decreased local oxygen tension are necessary for the development of an abdominal actinomycetoma. To progress, the infection habitually requires synergy with intestinal co-pathogens which act as “companion” bacteria, by producing toxins or enzymes, or by inhibiting the host immune response.^{1,2} As a result, perforated appendicitis is one of the most common causes of abdominal actinomycosis,^{2–5} the symptoms usually appearing several months or years after the initial infection.¹

The development of this infection is very indolent and is characterized by an intense local inflammatory response and a granulomatous abscess formation. Then, multiple contiguous abscess cavities form and extend, transgressing tissue planes. Internal and cutaneous fistulas are common. An extensive and indurated granulation zone is formed, composed of fibroblasts and collagen, often mimicking a neoplastic process.^{1,2}

Mild pyrexia and slight elevation in inflammatory values (leucocytosis, erythrocyte sedimentation rate, C-reactive protein) are frequent.² Otherwise, the symptoms are fairly non-specific, including change in bowel habits, nausea and vomiting, weight loss, pain and presence of a mass.¹ The investigation of the mass must include a neoplastic work-up, and generally may include radiology, echography, CT scan and MRI, in order to delineate the lesion and surrounding structures that may be implicated. There may also be a mass effect with compression of adjacent structures, such as hydronephrosis⁶ following compression of the ureter or neurovascular compromise.⁷

As this infection is rarely suspected, the diagnosis is usually made following biopsy or culture, which may take 1–2 weeks to identify actinomycosis¹ (Figs. 3 and 5). This bacterium secretes polysaccharides which provoke bacterial aggregation. The colony that is created is surrounded by polymorphonuclear neutrophils, forming the characteristic “sulfur granules”,¹ however present in only 50% of cases² (Fig. 4). It is particularly important to rule out the possibility of other bacteria that may mimic these granules, such as *Staphylococcus*, *Nocardia*, *Monosporium*, and *Cephalosporium*.^{1,2}

Fewer than 10% of actinomycetomas are diagnosed pre-operatively.² This demonstrates the large number of morbid surgeries that may be avoidable, as the correct treatment often consists of percutaneous abscess drainage in certain cases and long-term antibiotic therapy, usually 6–18 months, individualized according to clinical presentation.¹ In cases where open surgical intervention is necessary, debridement of all necrotic infected tissues may significantly decrease the bacterial count. The prognosis

is usually very good, however recurrence is frequent if the initial treatment is not complete.^{1,2}

It is very well documented through many case reports and case series that abdominal actinomycosis poses a great diagnostic challenge, often mimicking a neoplastic process, until identification of the gram-positive bacillus *A. israelii* following biopsy, culture, or more often after a morbid surgical tumor resection protocol.

In relation to previous appendicitis and the late complication of abdominal actinomycosis, Moosmayer⁸ described a patient who at 2 and a half years of age sustained an appendectomy for acute appendicitis, presented 10 years later with an acute abdomen, and CT revealed a right-sided mass adjacent to the psoas. Fine-needle aspirate revealed *A. israelii*.

Four articles have described actinomycetomas simulating various extra-abdominal soft-tissue neoplasms. Molnar⁹ described a case simulating retroperitoneal sarcoma or lymphoma after an acute abdomen with a left subcostal mass in a 36-year-old patient, diagnosed as actinomycosis after the second laparotomy. Radhi¹⁰ reported the case of a 7-year-old boy for whom biopsy suggested a retroperitoneal inflammatory pseudotumor, and actinomycosis was confirmed after surgical resection. This infection had spread from its pulmonary origin through diaphragmatic sinuses. Maradeix¹¹ described a 69-year-old woman with suspected liposarcoma of the buttock region, and biopsy confirmed actinomycosis. The infection occurred four years after acute appendicitis, likely through slow fistulisation. Kumar¹² described a particular case of a 14-year-old boy with an anterior distal thigh mass, superficial to the quadriceps, diagnosed as actinomycosis after surgical drainage. It is unclear how this patient sustained this infection in this very unusual location.

On the other hand, Björk¹³ described a child with rib rarefaction, pulmonary consolidation and pleural effusion with serologically confirmed actinomycosis, however the clinical course revealed a coincidental metastatic Ewing sarcoma of the lung with actinomycosis.

Therefore, this is to the best of our knowledge the first report of a para-iliac actinomycetoma having presented as a sarcoma, occurring as a late complication of appendicitis.

4. Conclusion

In the present case, we believe that the actinomycetoma was formed following the perforated appendicitis two years earlier, having dissected through the soft tissues from its point of origin in the ileo-caecal region, posteriorly toward the psoas, and then extending to each side of the right iliac wing. Thanks to the collaborative work of the pathologist and microbiologist, the diagnosis of actinomycosis was confirmed before surgery. We therefore performed an extensive surgical debridement of the actinomycetoma followed by long-term antibiotic treatment, instead of a surgical tumor resection protocol, for this patient who presented with a probable diagnosis of sarcoma.

While faced with a para-iliac soft-tissue mass, especially with a history of previous perforated appendicitis, one should consider the possibility of an abscess caused by pathogens of intestinal origin, including the gram-positive anaerobe *A. israelii*. General surgeons and orthopedic surgeons must be aware of the various clinical manifestations of abdominal actinomycosis, as it is often erroneously diagnosed as a neoplasm until late in the investigation or treatment. Failure to actively search for this pathogen, which is not detectable with routine staining techniques and may take up to 1–2 weeks to isolate, may lead to unnecessary morbid surgical procedures.

Conflict of interest

None declared.

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None declared.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

The three authors, Dr. Pierre Navarre, Dr. Marc H. Isler and Dr. Marie-Andrée Cantin, have contributed to all aspects of this paper, including data acquisition, manuscript writing and review. The authors approve the final version of this manuscript.

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