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A rare finding of *Actinomyces odontolyticus* abdominal actinomycosis presenting as abdominal wall and pericolic pseudotumoral mass

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A rare finding of *actinomyces odontolyticus* abdominal Actinomycosis presenting as abdominal wall and pericolic pseudotumoral mass

Abdominal actinomycosis refers to a rare chronic suppurative infectious occurrence, caused by filamentous Gram-positive microaerophilic and anaerobic bacteria Actinomyces, that may appear as an abdominal mass and/or abscess, feasibly mimicking a malignancy^{1,2}. Due to its rarity and unspecific clinical evidence, the majority of cases are diagnosed after tissue specimen. We hereby report a case of a 69-year-old patient with a one week worsening abdominal pain and swelling. A large tender palpable mass in the epigastric region was noted on physical exam. An ultrasound-guided drainage followed by a surgical excision approach became both a way to confirm the diagnosis and a therapeutic tool. Diagnosis of actinomycosis was made on histopathology and microbiology. Even though the incidence of actinomycosis has decreased, the abdominal presentation has been observed with increasing frequency³.

KEY WORDS: *Actinomycosis, Abdominal wall, Abdominal abscess, foreign-body reaction, Colonic neoplasms*

Introduction

Actinomycosis is an uncommon granulomatous and inflammatory disease produced by an opportunistic pathogen responsible for an invasive infection of the abdomen in 20% of actinomycotic infections⁴. *Actinomyces* bacteria belong to a strain that can be divided into six sub-groups of which *israelii* is the most common variety⁵. In our case, instead, we are presenting an uncommon case of infection related to *Actinomyces odontolyticus*, a commensal of the oral cavity, rarely pathogenic, able to cause infection only when it enters the tissue following a breach in the mucosal barrier. Since *Actinomyces* is an obligate anaerobe, it requires

many other types of bacteria to proliferate in order to destroy local tissue in any highly vascularized area and to replace it with a poorly irrigated granulation tissue which allows an anaerobic milieu⁶. Therefore, there is no specific radiological evidence of the disease. Later stages of the chronic suppurative infection that occurs in abdominopelvic actinomycosis may lead to the formation of an abdominal mass, with multiple abscesses and a severe inflammation that may involve the abdominal wall with fistulae or compromise the perianal area and other organs. It mostly affects middle-aged adults and is most common in males⁷. As the diagnosis is notoriously hard, it is often referred to as “the great pretender”, because of a broad spectrum of differential diagnosis such as inflammatory bowel disease, diverticulitis, tuberculosis, appendicitis or even malignancy⁸. Ileocecal segment, resulting from appendix perforation, is more frequently affected although stomach, liver, duodenum, gallbladder, colon, anorectal area and pelvic organs can be involved as well⁹. It has been reported how abdom-

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inal actinomycosis may occur following acute appendicitis, trauma, diverticulitis, abdominal surgery or in female patients with intrauterine devices¹⁰. This paper deals with a case of abdominal actinomycosis where the involvement of the transverse colon by the mass made a diagnosis of malignancy even more likely.

Case Report

A 69-year-old male, with a known past medical history of PTCA after AMI, emphysema, cholecystectomy, appendectomy and distal gastrectomy following perforated gastric ulcer, presented with a one week worsening epigastric pain and swelling. There were no gastrointestinal or urogenital related symptoms, no history of constitutional symptoms or any relevant change in bowel habit. Physical examination showed mild tenderness in the epigastric region where a hard and firm palpable mass was evident. On admission, laboratory tests revealed mild neutrophilic leukocytosis, together with lymphocytopenia, monocytosis, high RCP levels and normal carcinoembryonic antigen (CEA) and alfafetoprotein. Vital signs were unremarkable. Body temperature was 37.1-37.5°C during the first couple of days of hospitalization. Computerized tomography with contrast showed, between the transverse colon and the parietal peritoneum, an ill defined fluid density mass, measuring 7.5x4x3.5 cm, with polycyclic edges, parietal enhancement and an internal linear calcification, supposed to be a not better specified foreign body (Fig. 1).

The CT scan report was inconclusive: in consideration of the clinical presentation and the laboratory results, the lesion could be secondary to severe inflammatory reaction, while not ruling out a malignancy.

Colonoscopy was scheduled in order to identify any possible colonic lesion. However, the procedure was incomplete due to a colonic angle and substenosis of the splenic flexure.

His aspirin was held and an ultrasound-guided aspiration of the mass was performed. Cultures eventually grew *Actinomyces odontolyticus*. At exploratory laparotomy we found a mass of about 10 cm diameter surrounded by fibrous capsule, together with multiple cavities bordered with necrotic and suppurative material involving the abdominal wall till the subcutaneous tissue, tightly adhered to the transverse colon and cleavable at the same time (Fig. 2).

Intra-operative colonoscopy revealed a sessile white polyp similar to a granuloma of the transverse colon, right inside the bowel whose surface appeared overlaid by the mass (Fig. 3). During the endoscopic procedure, a polypoid lesion was removed at approximately 30 cm from the anal verge. There was no true invasion of either the transverse colon or the neighbouring structures. No bowel resection was necessary.

Therefore, en bloc excision of the mass and abundant intra-peritoneal lavage were performed.

Microscopic examination of the surgical specimen revealed cutaneous fragments together with muscle, fibrous and adipose tissues combined with steatonecrosis and acute suppurative inflammation, associated to rare giant cells englobing foreign round bodies birefringent under polarized light (Fig. 4 and 5).

The patient recalled to have the habit of flossing his teeth. Eventually, we believe he may have accidentally swollen part of the thread, responsible for conveying bacteria from the oral cavity through the gastrointestinal tract with subsequent mucosal barrier breach.

Pathologist included fragments of the braided dental



Fig. 1: CT scan imaging showed fluid density intraperitoneal mass, measuring 7.5x4x3.5 cm, with polycyclic edges and parietal enhancement. The red arrows in the axial and sagittal sections show the internal linear calcification, supposed to be a foreign body.

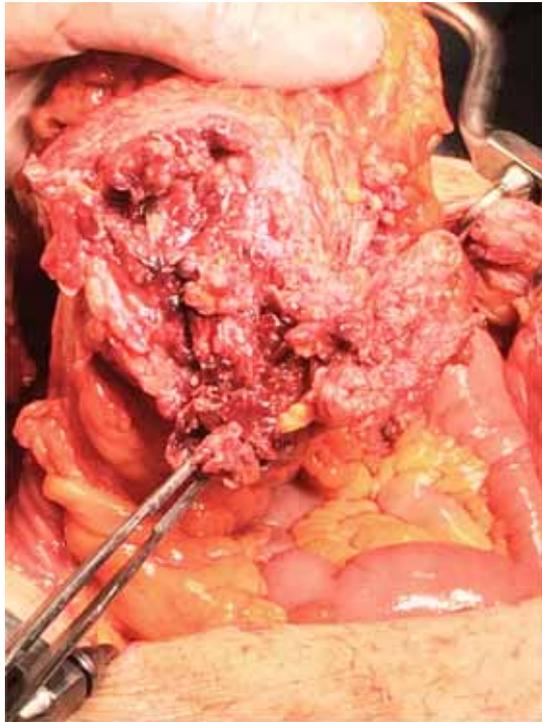


Fig. 2: Abdominal mass surrounded by fibrous capsule together with multiple cavities bordered with necrotic and suppurative material involving the abdominal wall till the subcutaneous tissue, tightly adhered to the transverse colon and cleavable at the same time.



Fig. 3: Colonic mucosa below the abscess lesion as it appeared at intraoperative colonoscopy.

floss, similar to the ones revealed in our specimen, inside a colonic human tissue in order to spot the signs and characteristics of the foreign-body reaction. Foreign bodies may provoke chronic inflammation whose pattern depends on the kind of tissue involved. However, the mechanical and enzymatic digestion may have altered the specific pattern of the material; therefore, we were able

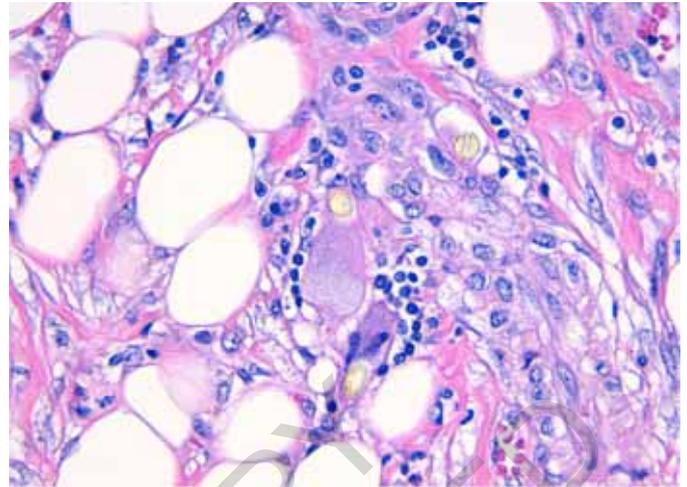


Fig. 4: Histopathology of the transverse colon specimen: fibrous and adipose tissues and inflammatory foci, associated to three likely braided fragments of foreign bodies engulfed by giant cells (haematoxylin-eosin stain 400x).

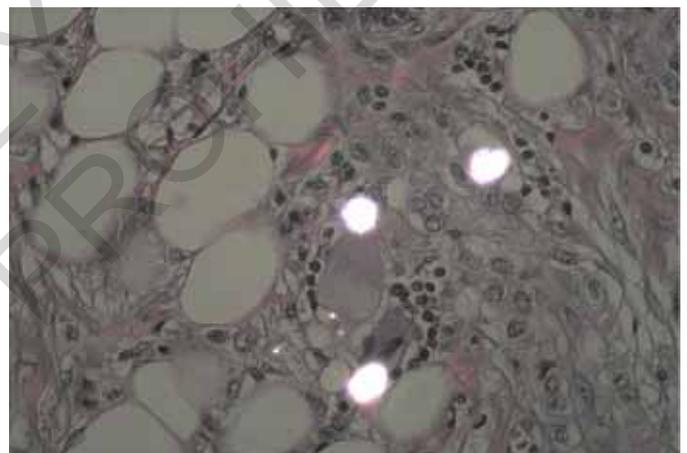


Fig. 5: Histological examination of the removed transverse colon specimen: birefringent foreign bodies incorporated by giant cells as part of a severe inflammatory reaction. (haematoxylin-eosin stain 400x).

to observe a resembling pattern due to the fact that the exact one, matching the histological features of our specimen, is hardly reproducible.

During recovery, Odontostomatology was called for evaluation and a severe status of periodontal disease was noted without any sign of acute inflammatory distress. Echocardiography excluded any evidence of endocardial vegetations.

The removed polypoid lesion was confirmed to be a benign one.

The post-operative course was unremarkable, except for a mild respiratory distress due to a bacteriological pneumonia in a patient with a severe anamnestic cigarette smoking related COPD.

He was started on high dose of intravenous piperacillin-tazobactam with appropriate response and then transi-

tioned to oral amoxicillin/clavulanic acid for about 6 months after being discharged from the hospital. Both early outpatient follow-up and 30-day outcome demonstrated excellent recovery.

Discussion

The overall incidence of registered cases of actinomycosis is decreasing, while pelvic and abdominal actinomycosis, however, are increasing³. Actinomyces species are common commensals of the oral cavity as well as of the gastrointestinal and urinary tracts. Hence, the three main clinical presentations of the disease are cervicofacial, thoracic and abdominopelvic actinomycosis¹¹. Human beings are considered the natural reservoir. Giving the nonvirulent nature of the bacteria, mucosal barrier disruption is mandatory in order to allow the progression of the infection and the involvement of surrounding tissues. Therefore, it can be clinically misdiagnosed as malignancy^{8,11}. Abdominal actinomycosis is one of the greatest challenges for diagnosis and reaches about 20% of the overall cases with its indolent and unspecific presentation, mimicking malignancy, inflammatory bowel disease and tuberculosis^{8,12}. Less than 10% of cases are diagnosed pre-operatively¹³. Recent surgery, appendicitis, diverticulitis, IUD, intraperitoneal spillage of gallstones during laparoscopic cholecystectomy, neoplastic processes or even foreign bodies may be considered among predisposing factors of the disease¹⁴. Most commonly, abdominal actinomycosis occurs in the ileocecal segment and rarely in ascending, left side and transverse colon^{15,16}. Despite the excellent response to antibiotics, full recovery is rarely achieved without surgery, which remains a mainstay of the therapy for the drainage of abdominal abscesses and the treatment of fistulae, intestinal obstructions and abdominal masses.

As in our experience, patients may have an unspecific clinical presentation, including low-grade pyrexia, mild abdominal pain and an abdominal mass, which can be identified through CT scans without any distinctive diagnostic feature. However, preoperative diagnosis may be aided by radiological-guided aspiration of the lesion and following collection of bacteriological specimens. As a matter of fact, in our case, cultures eventually grew Actinomyces odontolyticus. Moreover, radiological scans are likely to contribute to the definition of the anatomic location of the lesion and its extension. Intraoperative endoscopy can help to exclude intraluminal macroscopic colonic lesions. Surgery guides differential diagnosis with other diseases and removes the abdominal lesions, providing a pathological specimen. Hence, histopathological examination, in our experience, revealed a severe suppurative inflammation associated to giant cells, incorporating fragments of foreign bodies that we believe to be plastic material, responsible for conveying bacteria from the oral cavity through the gastrointestinal tract

and leading to the mucosal barrier breach. Other pathological evaluations describe the characteristic sulfur granules or unspecific signs of chronic granulomatous infection compatible with actinomycosis.

Conclusions

Abdominal actinomycosis is a rare indolent infectious disease that has to be included in a wide number of differential diagnosis¹⁷. Clinical and imaging findings are likely to suggest malignancy. Therefore, tissue and bacteriological specimens are necessary to establish the right diagnosis. Combined medical and surgical treatment is usually effective in about 90% of cases¹⁸. Nevertheless, delay in diagnosis of actinomycosis may lead to a complicated course, recurrence or even death. Increased awareness of the disease will encourage the index of suspicion and possibly lead to a preoperative diagnosis, avoiding unnecessary demolitive surgical treatments and non-effective antibiotic therapies¹⁹.

Riassunto

L'Actinomicosi rappresenta un processo infiammatorio e granulomatoso cronico prodotto da un patogeno opportunisto responsabile nel 20% dei casi di un severo processo infettivo addominale. L'incidenza globale di casi registrati di actinomicosi risulta in diminuzione, mentre l'interessamento pelvico ed addominale della patologia è in aumento. Stadi avanzati del quadro cronico suppurativo che contraddistingue l'actinomicosi addominopelvica possono manifestarsi quali massa addominale con multiple cavità ascessuali ed un severo quadro infettivo che può arrivare a coinvolgere la parete addominale con formazione di fistole o compromettere il perineo ed altri organi limitrofi. Date la rarità e l'aspecificità nelle evidenze clinico-radiologiche di questa patologia, la gran parte dei casi viene identificata grazie all'analisi istopatologica e microbiologica culturale. Nella nostra esperienza riportiamo il caso di un paziente di 69 anni affetto da dolore addominale ingravescente, associato all'obiettività clinica di massa palpabile epigastrica ed a quella radiologica di neoformazione contenente un presunto corpo estraneo e coinvolgente il colon trasverso. Il quadro clinico lasciava spazio ad un ampio spettro di diagnosi differenziali compreso il sospetto di malignità. L'aspirazione eco-guidata della massa ha rivelato l'eccezionale presenza in sede addominale di colonie di Actinomyces odontolyticus, noto commensale del cavo orale. Il paziente è stato sottoposto ad asportazione en bloc della massa che, come confermato anche mediante endoscopia intraoperatoria, non presentava franchi aspetti invasivi verso gli organi limitrofi; la diagnosi è stata ottenuta mediante analisi istologica e batteriologica. Pensiamo che il paziente, solito all'utilizzo quotidiano di

filo interdente, possa averne incidentalmente ingerito una minima parte che è servita quale veicolo per la migrazione del batterio commensale orale sino alla sede colica. L'inclusione sperimentale di frammenti plastici di filo interdente ha rivelato importanti somiglianze con il materiale estraneo riscontrato nel campione istologico del paziente. Il trattamento chirurgico insieme con la somministrazione di alti dosaggi antibiotici hanno consentito la risoluzione del quadro clinico. Spesso i reperti clinico-radiologici di questa patologia fanno supporre una eziologia maligna, per cui l'analisi istologica e microbiologica colturale divengono mandatorie per raggiungere una corretta diagnosi differenziale e definire il trattamento più adeguato, soprattutto alla luce delle severe complicanze che altrimenti potrebbero derivarne. Una miglior consapevolezza della patologia deve incoraggiare il tasso di sospetto e possibilmente guidare verso una diagnosi pre-operatoria nell'ottica di definire la strategia di trattamento più opportuna.

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