ACTINOMYCOTIC ILEAL PERFORATION: A CASE REPORT AND LITERATURE REVIEW

LEONARDO DELOGU1, PANAGIOTIS PALIOGIANNIS3, ALBERTO FARA1, ANTONIO COSSU3, FABRIZIO SCOGNAMILLO2, FEDERICO ATTENE2

1Operative Unit of Surgery, Alghero Civil Hospital, Alghero, Italy - 2Department of Surgical, Microsurgical and Medical Sciences, University of Sassari, Sassari, Italy - 3 Faculty of Medicine and Surgery, University of Sassari, Sassari, Italy

[Perforazione ileale actinomycotica: case report e revisione della letteratura]

ABSTRACT

Human actinomycosis is a chronic, slowly evolving infection caused generally by Actinomyces israelii. Diagnosis of abdominal actinomycosis is seldom made preoperatively due to the lack of specific clinical, laboratory and radiological features. This makes the clinical management challenging, furthermore considering the aggressiveness of the infection that often mimics a malignant mass or causes multiple abscesses and bowel perforation. We present here a case of ileal perforation due to Actinomyces israelii infection and a review of the current literature. An 86-year-old Italian woman with a painful 5 cm in maximum diameter hypogastric swelling was referred to our Unit. Blood tests revealed a mild leukocytosis. Abdominal computed tomography showed a 4 x 5 cm subcutaneous mass with unclear limits in correspondence of the infraumbilical region, with a low enhancement central area and diffusely enhanced peripheral limits. On laparotomy a great amount of purulent material was drained and a perforation of the ileum was evidenced. An ileal resection and a latero-lateral anastomosis was performed. The histological examination of the specimens evidenced an infection by Actinomyces israelii. The postoperative course of the patient was uneventful. Antibiotic therapy with penicillin was started once histological results were obtained. Follow up was performed 3 months after surgery; no recurrences or other complications were observed.

Key words: Actinomyces israelii, ileum, perforation, surgery, abscess, acute abdomen.

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Introduction

Actinomycosis is a chronic infection, with both suppurative and granulomatous features, caused by Actinomyces bacteria. The most common infection in humans is caused by Actinomyces israelii and rarely involves the gastrointestinal tract, especially the ileum. Diagnosis of abdominal actinomycosis is seldom made preoperatively due to the lack of specific clinical signs and symptoms, laboratory and imaging features and difficulty to evidence and isolate the microbe[1, 2]. This makes the clinical management and the choice of the appropriate therapeutic strategy challenging, furthermore considering the aggressiveness of the infection that often mimics a malignant mass or causes multiple abscesses and bowel perforation. We present in this article a rare case of ileal perforation due to Actinomyces israelii infection and a review of the current literature.

Case presentation

An 86-year-old Italian woman with a painful hypogastric swelling was referred to our Unit. The patient had not a family history of colorectal cancer or hereditary intestinal diseases, as well as personal history of previous abdominal surgery or intrauterine device (IUD). On physical examination the maximum diameter of the lump was approximately 5 cm and could not be reduced in the abdomen; no skin alterations were observed. A mild tenderness was evidenced, as well as regular bowel motility. The patient was pyrexic and did not referred
weight loss or alterations of the bowel habits. Blood tests revealed mild leukocytosis (11.7x10^3/μL, normal range: 5.2-10.5x10^3/μL) with slightly increased neutrophil count (8.8x10^3/μL, normal range: 1.9-8x10^3/μL); all the remaining hemocromocytometric parameters were within normal ranges, as well as the indices of hepatic and renal function.

An abdominal computed tomography (CT) was subsequently performed. It showed a 5 x 4 cm subcutaneous mass with unclear limits in correspondence of the infraumbilical region, characterized by a low enhancement central area and diffusely enhanced peripheral limits. The lesion resembled an inflammatory collection involving surrounding tissues. No free air in the abdomen was found. Routine preoperative evaluation was completed with chest X-ray and electrocardiography and an explorative laparotomy was performed.

The abscess mass was ruptured during laparotomy and a great amount of purulent material was drained (Figure 1) and samples were sent for microbiological, pathological and biochemical examination. Furthermore, the mass involved part of the ileum, cecum, omentum, and abdominal fasciae with numerous adhesions. Careful adhesiolysis was performed revealing a perforation of the ileum involved in the inflammatory process (Figure 2). This part of the ileum was subsequently resected, together with the inflamed part of the omentum and the abdominal fasciae. A latero-lateral ileal anastomosis was performed using a linear stapler and a drain tube was placed in the recto-uterine space of Douglas. Finally, the abdominal cavity was carefully washed and the abdominal wall reconstructed.

The histological examination of the specimens evidenced a mass characterized by intense inflammation with high amounts of polymorphonuclear leukocytes, fibroblasts and foamy histiocytes, vascular congestion, extravasation, and ialynosis. Fibrin deposits and hemorrhagic areas with multiple abscessual foci were evidenced in the ileal wall. Actinomyces Israelii was detected by p-aminosalicylic acid (PAS) staining method.

The postoperative course of the patient was uneventful. Antibiotic therapy with penicillin was started once histological and microbiological results were obtained. The patient was discharged nine days after surgery. Follow up was performed 3 months after the surgical operation with clinical and ultrasonographic evaluation; no recurrences or other complications were observed.

**Discussion**

Actinomycosis is a chronic infection, with both suppurative and granulomatous features, caused by Actinomyces bacteria. Human disease is usually determined by Actinomyces Israelii, a gram-positive anaerobic bacterium which is a commensal in several mucosae, especially the oral cavity, the gastrointestinal tract and the female genital tract. Male to female ratios in literature are discordant, but it seems that actinomycosis is more common in middle-age male individuals. The site of infection is most commonly the cervicofacial region (60%-40%), followed by the gastrointestinal tract (20%) and the thorax (15%). The gastrointestinal lesions usually involve the cecum, particularly the ileo-cecal region, while the descending colon is rarely affected. Furthermore, pelvic infections have been reported in several studies.

A disruption of the mucosal barrier is necessary for Actinomyces israelii to become pathogenous; events that can potentially cause mucosal damage like gastrointestinal surgery with or without appendicitis, diverticulitis, inflammatory ...
bowel disease, trauma, neoplasia and foreign bodies are risk factors for actinomycosis. Furthermore, longstanding IUDs have been reported to be related to higher incidences of pelvic actinomycosis\(^\text{[4]}\). Other predisposing conditions seem to be the administration of steroids, diabetes and immunodeficiency states\(^\text{[1,3]}\). In approximately half of the cases, no risk factors for mucosal disruption are found, as occurred in our case\(^\text{[2]}\). The mucosal damage might occur several years before the clinical manifestation of the disease, as actinomycosis is generally a slowly progressing inflammation.

Actinomycosis is rarely diagnosed prior to surgery (less than 10% of cases)\(^\text{[5,6]}\). Only few nonspecific clinical signs and symptoms characterize generally the disease, like fever, pain, nausea, vomiting, weight loss, diarrhea or constipation\(^\text{[1,8]}\). In our case the patient presented only a palpable mass in the hypogastric area and minor tenderness, which made the diagnostic process challenging. Perforation, if present, can produce serious complications and clinical manifestations, like acute abdomen, in relation to the site and entity of the perforative event, as well as to factors related to the patient’s general conditions, underlying diseases and the presence of one or more of the above-mentioned risk factors. Furthermore, no specific laboratory diagnostic markers for actinomycosis are available; leukocytosis, mild anemia and increase in inflammation indices are commonly found. Only a mild neutrophil leukocytosis was evidenced in our patient.

The role of imaging in the diagnosis of abdominal actinomycosis is important, even if no specific radiological features exist. CT scan can evidence several signs, like bowel wall thickening, disruption of mucosal folds, lumen strictures, presence of sinuses and/or intra-abdominal masses and collections. These findings are very similar to other acute or chronic inflammatory diseases of the gastrointestinal tract like Crohn’s disease, appendicitis, malignant tumors, tuberculosis and others. A study performed by Lee et al. evidenced that bowel wall thickening is concentric in most cases of actinomycosis, and it is generally adjacent to a solid, cystic or abscessual mass\(^\text{[2]}\). Actinomyces israelii have a natural propensity to cross the anatomical planes and invade adjacent tissues, sometimes mimicking a malignant neoplasia. Infiltration of adjacent organs and involvement of the abdominal wall fasciae are common findings in patients with chronic actinomycosis. Less commonly the infections involves the retroperitoneal space. Despite the relatively frequent occurrence of intestinal perforation in patients with abdominal actinomycosis, the peritoneal spread of the infection is rare, since the bacterium requires tissue contiguity to evolve. In addition, lymphatic dissemination seems to be exceptional, whereas blood stream dissemination is possible but rare. Computed tomography scan is useful for establishing the location, extent and possibly the nature of the disease. It is also helpful in the postoperative follow up, in order to evaluate the effectiveness of the antibiotic therapy and prevent recurrences.

Other useful diagnostic procedures include ultrasonography and barium-contrast radiography. Ultrasonography may be employed for transcutaneous drainage or biopsy, as reported by Schmidt et al.\(^\text{[13]}\). Furthermore, Lee et al. reported a case of diagnosis of pelvic actinomycosis by CT scan-guided transperietal biopsy\(^\text{[44]}\). As for barium study, the most common radiological signs of small intestine actinomycosis include mural invasion, stricture, mass effect with lumen narrowing, and thickened mucosal folds\(^\text{[2]}\). The clinical and endoscopic evaluation of the genital, intestinal and urinary tract may be useful to exclude neoplasms or other inflammatory conditions\(^\text{[2,3]}\).

Given the lack of specific clinical, laboratory and radiological features, preoperative diagnosis generally requires drainage or biopsy, followed by isolation, culture or microscopy detection of the microbe. Unfortunately, isolation of Actinomyces israelii is not easy as it aggregates in clusters scattered in different foci. Particular measures should be followed for bacterial culture, as the pathogen is a slow growing anaerobic organism, which needs up to 10 days to be evidenced. Histopathology often shows sulphur granules in the specimens, but they are not pathognomonic of actinomycosis: nocardiosis, streptomycosis, chromomycosis, botryomycosis and eumycetoma may present sulphur granules\(^\text{[18]}\). Direct recognition of Actinomyces species using haematoxylin and eosin staining can be challenging. Special stains, like Brown-Benn, Mc Callen-Goodpasture, Gomori methenamine silver or PAS can be employed. The latter method was used in our case.

The treatment of abdominal actinomycosis essentially consists in antibiotic therapy and surgery and resolves the disease in up to 90% of cases\(^\text{[3]}\). The therapeutic strategy must be tailored considering the site and the entity of the infection, the clini-
Surgery generally consists in an explorative laparotomy, adhesiolysis, removal of the abscess and cleaning of the abdominal cavity. Intestinal resections and anastomosis or Hartmann’s procedures are generally required in cases of intestinal perforation, as occurred in our patient. Larger resections, involving organs of the genital tract or other viscera are exceptional.

Conclusions

Human actinomycosis is a chronic, slow growing infection, generally caused by the Actinomyces israelii gender. Preoperative diagnosis of abdominal actinomycosis is rare because of the lack of peculiar clinical, laboratory and imaging features. This makes diagnosis challenging, given the difficulty to perform biopsies and to isolate the bacterium. CT scan of the abdomen is essential for staging the disease and plan the therapeutic strategy, which generally includes antibiotics and surgery. The clinical conditions of the patient, eventual comorbidities, past surgical operations and the site and entity of the infection must be taken into account in order to decide the most appropriate therapeutic approach.

References

11) Lee YK, Bae JM, Park YJ, Park SY, Jung SY. Pelvic actinomycosis with hydrenephrosis and colon stricture simulating an advanced ovarian cancer. J Gynecol Oncol 2008; 19; 154.