

Actinomycosis of the small intestine: A rare cause of perforation

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ABSTRACT

Abdominal actinomycosis is extremely rare. We report a case of actinomycosis of the small intestine presenting as a perforation and requiring subsequent laparotomy and small bowel resection. Such a description of actinomycosis has not previously been described in the literature.

Key words: Actinomycosis, perforation, small bowel

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INTRODUCTION

Actinomycosis is an extremely uncommon condition, most often caused by the gram-positive anaerobic bacterium, *Actinomyces israelii*; a component of the human oral and gastrointestinal flora. The cervicofacial region is the most common site of the disease followed by the abdominopelvic region, with the bacteria initiating an inflammatory response in the affected area resulting in sinuses, abscesses and fistulae.^[1] We report an extremely rare case of actinomycosis of the small intestine presenting as a perforation. This has not been previously described in the literature.

CASE REPORT

A 92 year-old female with no significant co-morbidity presented as an emergency with a 24-hour history of sudden, severe, central abdominal pain with associated anorexia, vomiting and diarrhea. This was on a background of a two-year history of vague abdominal discomfort and weight loss. On examination, she appeared dehydrated and distressed yet apyrexial and hemodynamically stable. She displayed upper abdominal peritonism. Erect chest X-ray and supine abdominal X-ray were unremarkable and blood

tests revealed a neutrophilia and elevated C-reactive protein (CRP).

Subsequent computed tomography (CT) scan revealed findings consistent with perforation of a hollow viscus.

Following lengthy discussion with the patient and next of kin regarding the inherent risks of surgery, exploratory laparotomy was performed. Laparotomy revealed a large amount of purulent intraperitoneal fluid and an isolated perforation of the distal ileum with adjacent inflammatory change. A small bowel resection of 10 cm of the terminal ileum was performed with hand-sewn, serosubmucosal anastomosis.

Subsequent histological analysis of the specimen revealed a transmural defect of the distal ileum consistent with perforation with a patchy fibrinous coat. On opening the bowel, multiple large ulcers were present which, on microscopy, revealed a complete loss of ileal mucosa with extensive ulceration and transmural inflammation. Sections through the perforation showed a perforation track with peritonitis and actinomycotic colonies lying both within the track and in the wall of the track. There were also large numbers of actinomycotic colonies present within the surface inflammatory exudate. These appearances were all felt to be in keeping with actinomycotic ileitis with perforation.

Long-term penicillin therapy was initiated postoperatively and the patient made an unspectacular recovery.

DISCUSSION

Nontraumatic perforation of the small intestine is extremely rare in western countries, with the etiology most commonly ascribed to Crohn's disease, strangulation, postoperative complications and malignancy.^[1] Actinomycosis of the small intestine presenting as a perforation has not been reported previously in the literature. In a review by Clinton *et al.*, the incidence of abdominal actinomycosis alone was found to be between 1 in 119,000 and 1 in 400,000 per year.

Actinomycosis is a chronic disease characterized by abscess formation, tissue fibrosis, draining sinuses and ulcers. It is caused by the filamentous, gram-positive anaerobic or microaerophilic bacterial species of the genus *Actinomyces*-the most common in humans being *Actinomyces israeli*.^[2] The pathogenic *Actinomyces* species do not exist freely but are commensals in the oral cavity and gastrointestinal tract and to a lesser extent, in the female pelvis. The portal of entry is typically a break in the mucosa of the gastrointestinal tract anywhere from the mouth to the rectum, occurring due to bacterial suppurative, diverticulitis, appendicitis, surgery or trauma.^[3,4] Implantation of the *Actinomyces* species into damaged tissue eventually leads to the development of chronic, indurated, suppurative infection often with draining sinuses and fibrosis. Three clinical forms of actinomycosis account for the majority of infections in humans-cervicofacial (50-70%), thoracic (15-20%) and abdominopelvic (10-20%).^[5] In the above case of abdominal actinomycosis, the infection and inflammatory response ultimately led to perforation of the distal ileum and due to this observation, we will concentrate only on abdominal actinomycosis.

Abdominal actinomycosis may be the most indolent and latent of all the clinical forms of the disease. Diagnosis may be delayed months to years after the precipitating event or as in the above case, made only after a catastrophic complication such as perforation. There is a predilection for the ileocecal region of the bowel and thus, it can easily mimic colonic adenocarcinoma, intestinal tuberculosis, chronic appendicitis, regional enteritis or amoeba.^[6] Gastric, perigastric, hepatic, splenic and renal involvement is uncommon but the species may reach these viscera through direct extension from the bowel or an intraabdominal site or via seeding through the portal vein.^[7] Abdominal actinomycosis may spread into the pelvis or alternatively, primary involvement of pelvic structures may arise in association with infection of intrauterine contraceptive devices^[8] (IUCDs).

In the majority of cases, the main difficulty is in making a definitive diagnosis without the need for surgery. CT

scanning is the most helpful diagnostic modality-the appearance of a contrast-enhancing multicystic lesion often suggesting actinomycosis.^[9] If the presenting symptoms allow time for investigation, tissue biopsies or samples from aspirated pus often show the frequently described "sulphur granules". These clumps of filamentous actinomycete microcolonies surrounding neutrophils may not always be pathognomic of actinomycosis.

Treatment of abdominal actinomycosis is dependent on both the extent of the disease and the health of the patient. For uncomplicated cases, the standard medical treatment of choice would be long-term penicillin. A combined medical-surgical approach is often necessary in order to treat complex actinomycosis. Surgery is indicated for resection of necrotic tissue, excision of sinus tracts or perforated segments and drainage of abscesses. Although surgery facilitates recovery, it is not curative on its own and additional long-term medical therapy is necessary for a good recovery.

This case is an extremely rare form of presentation of the disease and there are no other reported cases of actinomycosis of the small intestine presenting as perforation. The vague history of weight loss and chronic, nonspecific, abdominal pain is certainly consistent with actinomycosis although one would normally expect to find an abdominal mass on clinical examination or CT scanning. Nevertheless, in this case where the patient presented with an acute abdomen with CT scanning revealing intraperitoneal free air, surgical intervention was necessary and ultimately successful, with long-term antimicrobials serving as an adjunct to surgery.

Actinomycosis occurring within the abdominal cavity is extremely rare. However, it is a diagnosis, which should be considered in patients presenting in an atypical fashion where their history and examination do not correlate with more common pathology. In the above case, surgical intervention and small bowel resection was necessary. However the more chronic form of the disease is often amenable to treatment with antibiotics, thus obviating the need for surgery.

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