Ileo-caecal actinomycosis : Report of a case simulating complicated inflammatory bowel disease

A. Postal[1], O. Detry[1], E. Louis[2], N. Hardy[3], J. Belaïche[2], N. Jacquet[1]

(1) Department of Abdominal Surgery ; (2) Department of Gastroenterology ; (3) Department of Pathology, University Hospital of Liège, CHU Sart Tilman B35, B-4000 Liège, Belgium.

Abstract

Abdominal actinomycosis is a rare infectious disease caused by *Actinomyces israelii*, a gram-positive anaerobic saprophyte germ that is a normal inhabitant of the upper intestinal tract in humans. *Actinomyces israelii* rarely cause abdominal infections or actinomycosis. Abdominal actinomysosis is characterised by fistulae and abscesses and may mimic cancer or inflammatory bowel disease. Abdominal actinomycosis is difficult to diagnose preoperatively, and often require surgical removal of the diseased tissue, allowing pathologists for giving the definitive diagnosis, revealed by characteristic "sulfur granules". The authors report herein the case of a 47-year-old man who presented with diarrhoea and abdominal pain. Abdominal computed tomography evoked complicated inflammatory bowel disease and surgical procedure was decided. Laparoscopic exploration did not provide further significant information, and laparotomy with diseased bowel resection was performed. Pathology demonstrated "sulfur granules" and allowed the diagnosis of abdominal actinomycosis. This case demonstrated that abdominal actinomycosis should be included in the differential diagnosis when computed tomography shows an infiltrative and inflammatory mass. (Acta gastroenterol. belg., **2001**, 64, **318-320**).

Key words : case report, review, actinomycosis, surgery, treatment.

Introduction

Abdominal actinomycosis is a rare disease caused by *Actinomyces israelii*, a gram-positive, anaerobic, filamentous saprophyte germ, that may be a normal component of the endogenous flora of the mouth and upper digestive tract. It may become pathogen only in presence of damaged mucosa or necrotic tissue. In this paper the authors report the case of a patient who suffered from abdominal actinomycosis simulating complicated inflammatory bowel disease. The patient required laparotomy with resection of the diseased tissue, and long-term antibiotherapy. This case demonstrated that abdominal actinomycosis should be considered in the differential diagnosis when computed tomography shows an infiltrative and inflammatory mass.

Case Report

A 47-year-old male patient presented with abdominal pain to the emergency department. His past medical history revealed surgical and radiological procedures for a cerebral angioma. He was treated with diphantoïn. He had no past abdominal medical history. The patient had suffered from mild fever for one week. The abdominal pain was diffuse but more intense in the right lower quadrant. Diarrhoea was noted. The clinical examination revealed a patient in good general condition, but slightly dehydrated. The body temperature was measured at 37.7 °C. The abdomen remained overall flexible, except in the right lower quadrant, where clear defence and rebound sign were present. The abdominal sounds were increased. Laboratory tests revealed an important inflammatory syndrome with a leucocytosis at 18,890 white blood cells by mm^3 (normal values : 4,700-10,300), C Reactive Protein at 274 mg/L (normal values : 0-6), fibrinogen at 6.72 g/L (normal values : 1.90-3.70). There was also a slight anaemia (haemoglobin 13.5 gr/100 mL ; normal values : 13.7-16.8). The hepatic tests were normal, as well as the pancreatic enzymes. The ions and the renal function were normal. The urinary sediment was normal. The patient underwent an abdominal radiography that did not show any evidence of occlusion or perforation. Abdominal ultrasound showed a thickening of the walls of the caecum and the right colon. The diagnoses of acute appendicitis, right colon diverticulitis or inflammatory bowel disease were evoked. An abdominal computed tomography showed a marked inflammatory process of the ileo-caeco- appendicular junction (Fig. 1). The last ileal loop was thickened, as well as the right mesenteric fossa. All these findings were consistent with the diagnosis of complicated Crohn's disease, with probable mesenteric abscess and/or fistula.

The patient underwent diagnostic laparoscopy but this procedure showed an impressive inflammatory magma including the last ileal loop and the caecum, and open resection was decided. Sixty centimetres of final ileon, and the ascending colon were resected with restoration of the bowel continuity with a manual end-to-side ileo-colic anastomosis. The exploration of the abdominal cavity did not show any other organic lesion.

The pathology analysis described multiple characteristic "sulfur granules" (Fig. 2), abscesses located on the level of the mesenteric axis, and a fistula originating from the appendix. Intestinal mucosa was normal, and consequently, Crohn's disease was excluded. Stool

Mailing correspondence to : Olivier Detry, Department of Abdominal Surgery, CHU Sart Tilman B35, B-4000 Liège, Belgium. E-mail : Oli.Detry@chu.ulg.ac.be.

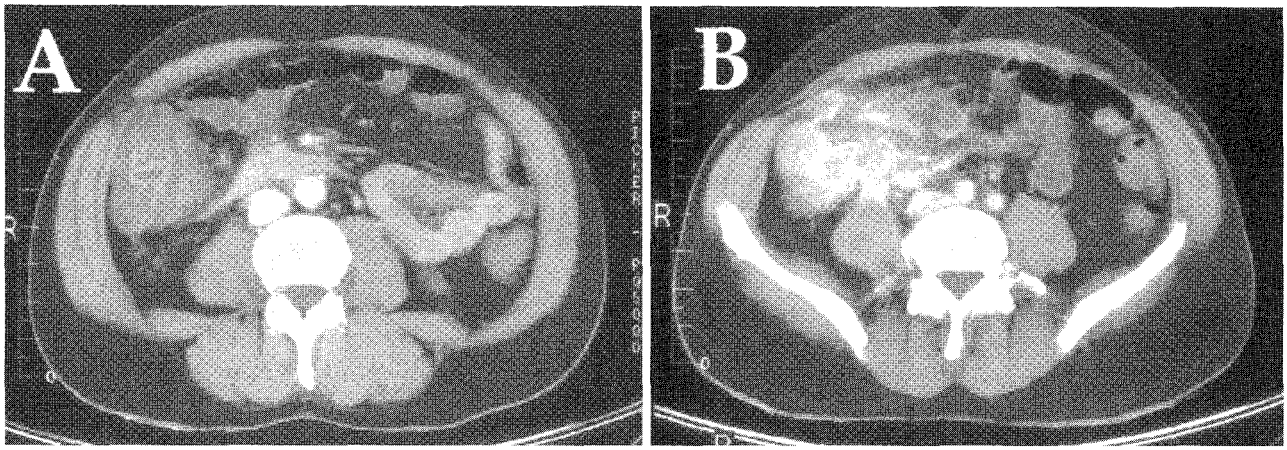

Fig. 1. — Computed tomography of the abdomen showing thickening of the right colon wall (Fig. 1A) and an inflammatory mass of ileocaecal junction, compatible with the diagnosis of complicated inflammatory bowel disease (Fig. 1B).

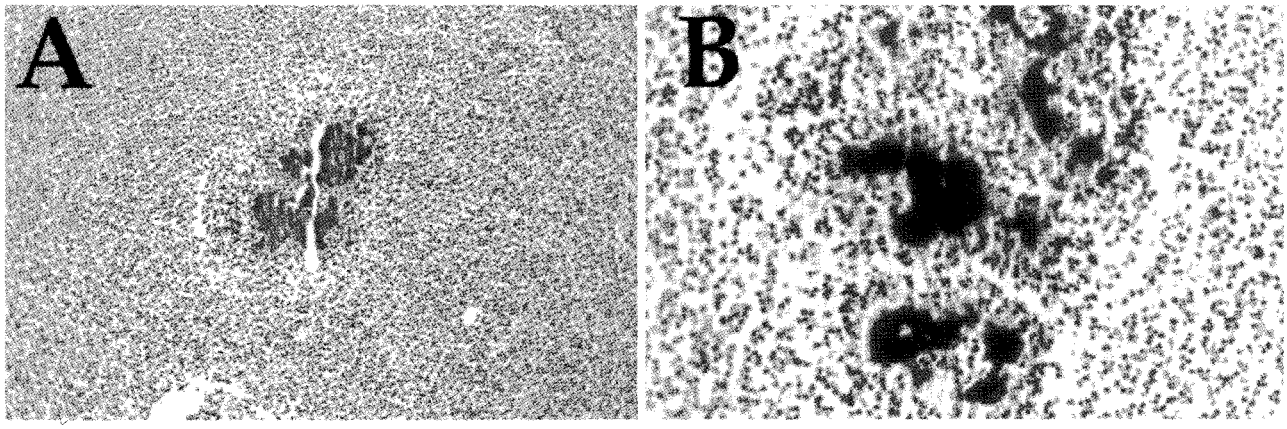

Fig. 2. — Pathology of the resected tissue (hematoxylin–eosin, 100 ×, Fig. 2A and 400 ×, Fig. 2B) showing the histologic aspect of "sulfur granules" surrounded by deteriorated polynuclear cells.

culture remained negative. Final diagnosis was abdominal actinomycosis. Antibiotherapy was initiated, with intravenous ceftriaxone (Rocéphine, Roche, Brussels, Belgium) (2 g/d, during 6 weeks) followed by oral amoxicillin (1,500 mg/d) (Clamoxyl, SmithKline Beecham, Genval, Belgium) during one year. Two years after the operation, the patient is asymptomatic.

Discussion

Human actinomycosis is rare infection due to an anaerobic, filamentous, gram-positive bacillus, *Actinomyces*. In humans, *Actinomyces israelii* is the usual pathogenic organism, but others may also be rarely found in human disease : *Actinomyces naeslundii, Actinomyces odontolyticus, Actinomyces viscosus, Actinomyces meyeri* and *Actinomyces propionica. Actinomyces bovis* were never found in human, and seem limited to animal pathology (1). Initially, *Actinomyces* was considered as a fungus. At present, it is completely acquired that this germ is an anaerobic gram-positive bacillus. The lack of a nuclear membrane, absence of chitin from cell walls, reproduction by fission, and most importantly, inhibition of growth by penicillin and insensitivity to amphotericin B classifie *Actinomyces* as bacteria and not fungi (1). Actinomycosis has a worldwide distribution, predominating in areas with poor standards for dental care. The incidence varies between 1/119,000 in the Netherlands and 1/40,000 in Cologne per year. It is three to four times more common in males than in females (1).

Actinomyces is a normal component of the endogenous flora of the human and animal oral cavities and upper digestive tract. They become pathogens only in presence of damaged or necrotic tissue. Actinomycosis can arise in various locations. The cervico- facial actinomycosis is the most frequent (50%). A chronic actinomycosis of the lower jaw may complicate tooth extraction. It is more rarely presented after a chronic dental abscess. The thoracic actinomycosis causes fistulae coming from consecutive pulmonary infiltrates secondary to the inhalation of oral commensal bacteria (1). Abdominal actinomycosis accounts for 20 to 23% of the human actinomycosis. Ten to 15% of human actinomycosis are rarer forms, involving hepatic, osseous, perianal (2), amygdalic, parotidic, lymphatic, testicular or

articular tissue. In abdominal actinomycosis, the appendix and ileocecal valve areas are the most common involved locations (1). *Actinomyces israelii* normally inhabits the colon, predominating in stagnation areas, *i.e.* the caecum and appendix. *Actinomyces* does not penetrate normal mucosa and requires mucosal injury to become pathogen. Predisposing factors include appendicitis, gastrointestinal perforations, previous surgery, foreign bodies, Crohn's disease or neoplasia. Once the organisms have penetrated the mucosa, spread by continuity seems to be the primary method of intraabdominal propagation. Lymphatic and hematogenous spread is uncommon.

Actinomycosis is a disease with chronic, slow evolution, characterised by the formation of fistulae with flow of granulomatous purulent material and by the presence of a granulomatous inflammatory infiltrate. After having crossed the intestinal mucosal membrane, the disease propagates gradually, with formations of multiple abscesses centred on *Actinomyces* colonies (1). The chronic infectious process reaches the neighbouring organs by local invasion. As the infection progresses, granulation tissue, fibrous tissue, multiple abscesses, and draining sinuses are formed. Clinically, abdominal actinomycosis is usually indolent with symptoms appearing one month to two years before definitive diagnosis. Associated findings include pain, weight loss, fever, leucocytosis, palpable mass, visible fistula tract, or fistulae. Abdominal actinomycosis may also cause entero- enteric, entero- vesical (3) or entero-cutaneous fistulae (4). Therefore, abdominal actinomycosis often imitates carcinoma, sarcoma, diverticular abscess, complicated inflammatory bowel disease, or abdominal tuberculosis. Diagnosis is determined preoperatively in fewer than 10% of the cases (5,6,7).

Definite diagnosis is generally established post-operatively by the pathology demonstrating *Actinomyces* colonies in the resected tissue and excluding cancer or Crohn's disease. The well recognised "sulfur granules" allow the diagnosis of the infection and may seldom be seen with the naked eye (1). The "sulfur granules" consist of agglomerate of *Actinomyces* colonies within a complex of polysaccharides and proteins (Fig. 2). These granules are of yellow-brown, and measure from 0.25 to 1 mm of diameter. Inflammatory granuloma may develop around these granules. They have three zones : a central abscess suppurated with deteriorated polynuclear cells ; a transition zone with varied inflammatory cells ; a peripheral zone consisting in a fibrosis whose thickness may vary according to the disease duration (1). Abdominal actinomycosis complicating Crohn's disease may be difficult to differentiate. The distinguishing histopathological features of secondary actinomycosis include multiloculated abscesses, sulfur granules, typical organisms seen with Gram stain, and rarely, necrotizing granulomata (8).

Preoperative diagnosis may be very difficult. Several factors contribute to the complexity of this diagnosis : the rarity of this disease makes that it is not considered in the differential diagnosis. *Actinomyces* is a strictly anaerobic bacterium whose growth in culture medium is difficult. *Actinomyces* is a saprophyte germ of the oral cavity and digestive tract, with possibility of false-positive culture. Radiological studies may contribute to the diagnosis but are not specific. Computed tomography seems to be helpful, demonstrating a solid mass with focal areas of attenuation or a cystic mass with a thickened wall that enhances with contrast infusion (9,10). The mass may be extra- or intra- luminal. It is "infiltrative" and may be associated with an intense desmoplastic reaction, making it difficult to differentiate from a malignant process or from perforated Crohn's disease. However, computed tomography in conjunction with fine needle aspiration may not only be diagnostic but also therapeutic (11).

Once the diagnosis is made, actinomycosis management is very successful. Frequently the treatment requires surgical resection for definitive diagnosis, but surgery may also be helpful as a therapeutic adjunct, permitting debridement of necrotic tissue and removal of persistent fistulae. Medical treatment is very effective in eradicating the disease, as *Actinomyces* is very sensitive to many antibiotics, amongst others to penicillin. However, because of the large amount of reactive fibrosis formed by the infection, it requires long-term therapy. A treatment with high dose of penicillin G, 10 to 20 million units per day for six to 12 months, is usually recommended (1).

References

1. CINTRON J.R., DEL PINTO A., DUARTE B., WOOD D. Abdominal Actinomycosis :report of two cases and review of the literature. *Dis. Colon Rectum*, 1996, **39** : 105-108.
2. MAST P., VEREECKEN L., VAN LOON C., HERMANS M. Actinomycosis ot the ano-rectum : a rare infection mimicking carcinomatosis. *Acta Chir. Belg.*, 1991, **91** : 150-154.
3. RICHARDS R.J., GRAYER D. Actinomycosis : a rare cause of vesicocolic fistula. *Am. J. Gastroenterol.*, 1989 ; **84** : 677-679.
4. KLAABORG K.E., KRONGORG O., OLSEN H. Entercutaneous fistulization due to Actinomyces odontolyticus : report of a case. *Dis. Colon Rectum*, 1985, **28** : 526-527.
5. MEYER P., NWARIAKU O., MCCLELLAND R.N., GIBBONS D., LEACH F., SAGALOWSKY A.I., SIMMANG C., JEYARAJAH D.R. Rare presentation of actinomycosis as an abdominal mass : report of a case. *Dis. Colon Rectum*, 2000, **43** : 872-875.
6. YEGUEZ J.F., MARTINEZ S.A., SANDS L.R., HELLINGER M.D. Pelvic actinomycosis presenting as malignant large bowel obstruction : a case report and a review of the literature. *Am. Surg.*, 2000, **66** : 85-90.
7. THOMPSON J.R., WATTS R., THOMPSON W.C. Actinomycetoma masquering as an abdominal neoplasm. *Dis. Colon Rectum*, 1982, **25** : 368-370.
8. MANLEY P.N., DRHU R. Actinomycosis complicating Crohn's disease. *Gastroenterology*, 1980, **79** : 934-937.
9. UCHIYAMA N., ISHIKAWA T., MIYAKAWA K., IINUMA G., NAKAJIMA H., USHIO K., YOKOTA T., AKASU T., SHIMODA T. Abdominal actinomycosis : barium enema and computed tomography findings. *J. Gastroenterol.*, 1997, **32** : 89-94.
10. HA H.K., LEE H.J., KIM H., RO H.J., PARK Y.H., CHA S.J., SHINN K.S. Abdominal Actinomycosis : CT findings in 10 patients. *Am. J. Roentgenol.*, 1993, **161** : 791-794.
11. GOLDWAG S., ABBITT P.L., WATTS B. Case report : percutaneous drainage of periappendiceal actinomycosis. *Clin. Radiol.*, 1991, **44** : 422-424.