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RESEARCH ARTICLE

ACTINOMYCOSIS MIMICKING A CROHN'S DISEASE

Omar Belkouchi¹, Omar El Malki², Lahssan Ifrine³, Abdelkader Belkouchi⁴, Lina Belkouchi⁵, Rachida Saouab⁶, Benayad Aourarh⁷, Aziz Aourarh⁸ and Jahid Ahmed⁹

1. Surgery Resident, Department of Surgery A, Ibn Sina Hospital, Mohammed V University, Faculty of Medicine and Pharmacy, Rabat, Morocco.
2. Surgery Professor, Department of Surgery A, Ibn Sina Hospital, Mohammed V University, Faculty of Medicine and Pharmacy, Rabat, Morocco.
3. Surgery Professor, Department of Surgery A, Ibn Sina Hospital, Mohammed V University, Faculty of medicine and Pharmacy, Rabat, Morocco.
4. Surgery Professor, Department of Surgery A, Ibn Sina Hospital, Mohammed V University, Faculty of medicine and Pharmacy, Rabat, Morocco.
5. Radiology Resident, Department of Radiology, Mohammed V Military Hospital, Mohammed V University, Faculty of Medicine and Pharmacy of Rabat, Morocco.
6. Radiology Professor, Department of Radiology, Mohammed V Military Hospital, Mohammed V University, Faculty of Medicine and Pharmacy of Rabat, Morocco.
7. Gastroenterology Resident, Department of Gastroenterology I, Mohammed V Military Hospital, Mohammed V University, Faculty of Medicine and Pharmacy of Rabat, Morocco.
8. Gastroenterology Professor, Department of Gastroenterology I, Mohammed V Military Hospital, Mohammed V University, Faculty of Medicine and Pharmacy of Rabat, Morocco.
9. Pathology Professor, Department of Pathology, Ibn Sina Hospital, Mohammed V University, Faculty of Medicine and Pharmacy, Rabat, Morocco.

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Abstract

Actinomycosis is a slowly progressive chronic granulomatous bacterial infection caused by an anaerobic gram-positive bacillus: Actinomyces Israeli. Thinking about this diagnosis in the right iliac fossa is uncommon as it usually shows through a clinical aspect of acute appendicitis, a Crohn's disease or even a tumor of the ileocolic region. We report the case of a young female patient admitted for an iliac fossa pain and a palpable mass, mimicking a Crohn's disease during laparoscopic exploration, only to conclude it's a retained diagnosis of actinomycosis, after doing an ileocecal resection and anatomopathological analysis. The purpose of this case report is to focus on the different aspects of this infection in the ileocecal region which can be mistaken at first hand that is only supposed to be treated medically by antibiotics for months, with a simple biopsy to confirm the diagnosis since surgery can be mutilating.

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Introduction:-

Actinomycosis is a chronic suppurative infection, secondary to a gram-positive bacillus: Actinomyces Israeli. The right iliac fossa localization is common; however, diagnosis remains uncertain, especially as it can be confused with

Corresponding Author:- Omar Belkouchi

Address:- Surgery Resident, Department of Surgery A, Ibn Sina Hospital, Mohammed V University, Faculty of Medicine and Pharmacy, Rabat, Morocco.

other inflammatory pathologies such as Crohn's disease, infectious (appendicular plastron or tuberculosis) or tumoral origin.

The specific diagnosis of actinomycosis is based on the anatomopathological analysis, through endoscopy biopsies or surgical biopsies and anaerobic pus cultures.

Imaging does not provide sufficient information or arguments to establish the diagnosis.

We report the case of a female patient admitted for right iliac fossa pain with a palpable mass, with an uncertain diagnosis, to whom an ileocecal resection was made by laparoscopy, with an actinomycosis diagnosis retained. Therefore, the surgical treatment was followed by long-term antibiotics.

Through this case report and literature review, we will show that cases in which actinomycosis is highly suspected, treatment can be based on the use of antibiotics alone for several months, and surgery should be reserved for other particular situations.

Case Description:

A 42-year-old woman, with no medical history, consulted for a right iliac fossa pain.

Symptoms started a month prior to admission, with local right iliac fossa pain, without any other associated symptoms such as vomiting or bowel disorders.

The patient was in a good health condition, with no signs of weight loss or fever. Her blood pressure was normal, her heart rate was at 70 beats/minute. Abdominal palpation revealed a painless abdominal swelling of the right iliac fossa, measuring about 5 cm in diameter.

Blood tests revealed a normal hemoglobin level of 13 g/dL (N=13-16g/dL), leukocytes at 10980/mm³(N= 4000-10000/mm³), with neutrophils at 7600/mm³ (N= 1500-8000/mm³), CRP (C-reactive protein) at 22 mg/L (Normal levels <5mg/L), creatinine at 4.5 mg/L (6-12mg/L), normal blood glucose levels at 1 g/L (N=0,7-1,1 g/L)), PT at 90% (N= 70-100%).An abdominal ultrasound and CT-scan revealed an inflammation of the ileocecal region that was predominant on the appendix associated with parietal fistula. Moreover, no ovarian mass was seen on the ultrasound. We only checked the tumor marker level of: CEA at 2 ng/mL (N<5ng/mL), and CA19-9 at 5 U/mL (N<37U/mL).Colonoscopy revealed normal colonic mucosa with extrinsic compression of the caecum that was very inflamed.The Bauhin's valve couldn't be crossed.Biopsy results of the colonic mucosa were inconclusive, showing no specific inflammation.

This is a young 42-year-old woman, hospitalized for a mass of the ileocecal region to which radiological imaging features led to a strong suspicion of either Crohn's disease or appendicular plastron.

Laparoscopic exploration showed some pelvic ascites that were translucent and yellow with a fairly inflamed right iliac fossa, a lot of adhesions to the abdominal wall and an appendix fistula. This macroscopic aspect resembles Crohn's disease; therefore, we performed an ileocecal resection, removing part of the abdominal wall as an en-bloc resection. Restoration of intestinal continuity was ensured by a manual, latero-lateral ileocecal anastomosis via a Pfannenstiel incision.

The anatomopathological analysis of the surgical specimen concluded that it is actinomycosis due to yellow sulfur granules found in the specimen.

The patient had parenteral ampicillin for 5 days, then we switched to an enteral prescription at a rate of 3 g per day, for 4 months. The follow up was good and no complications were noted.

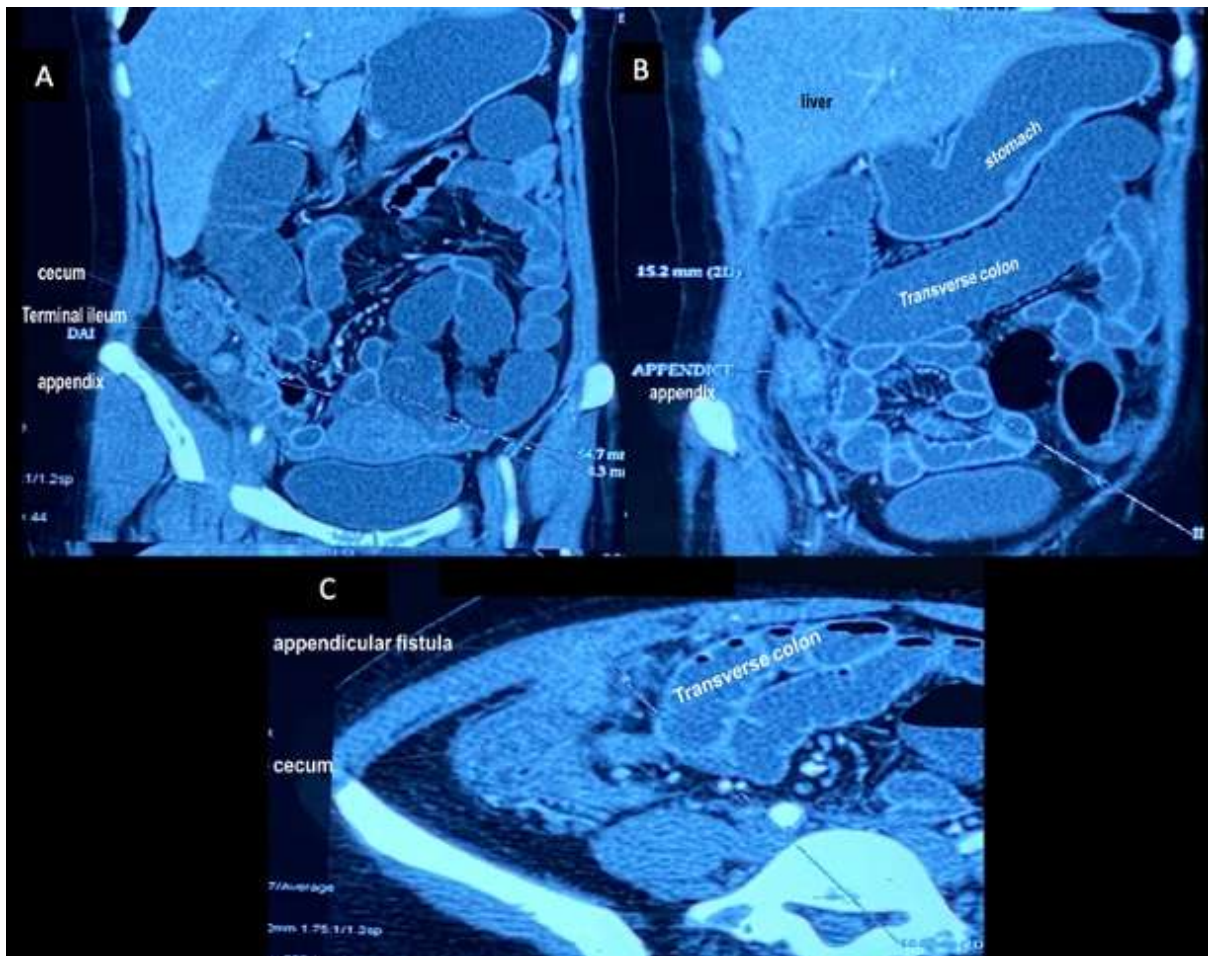


Figure 1:- Abdominal enhanced CT scan in coronal (A,B) and axial (C) images showing an inflammatory aspect of the ileo-ceco-appendix region (A and B) with a parietal fistula forming from the appendix (C).

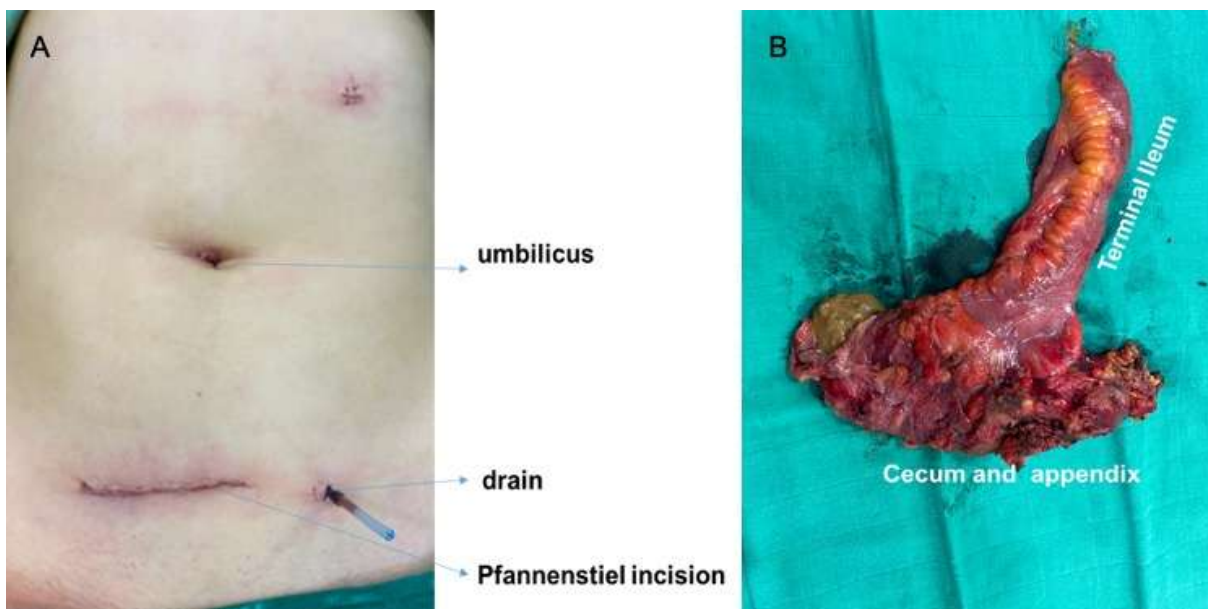


Figure 2:- Intra operative images showing the position of trocars and incision (A) and the surgical specimen (B).

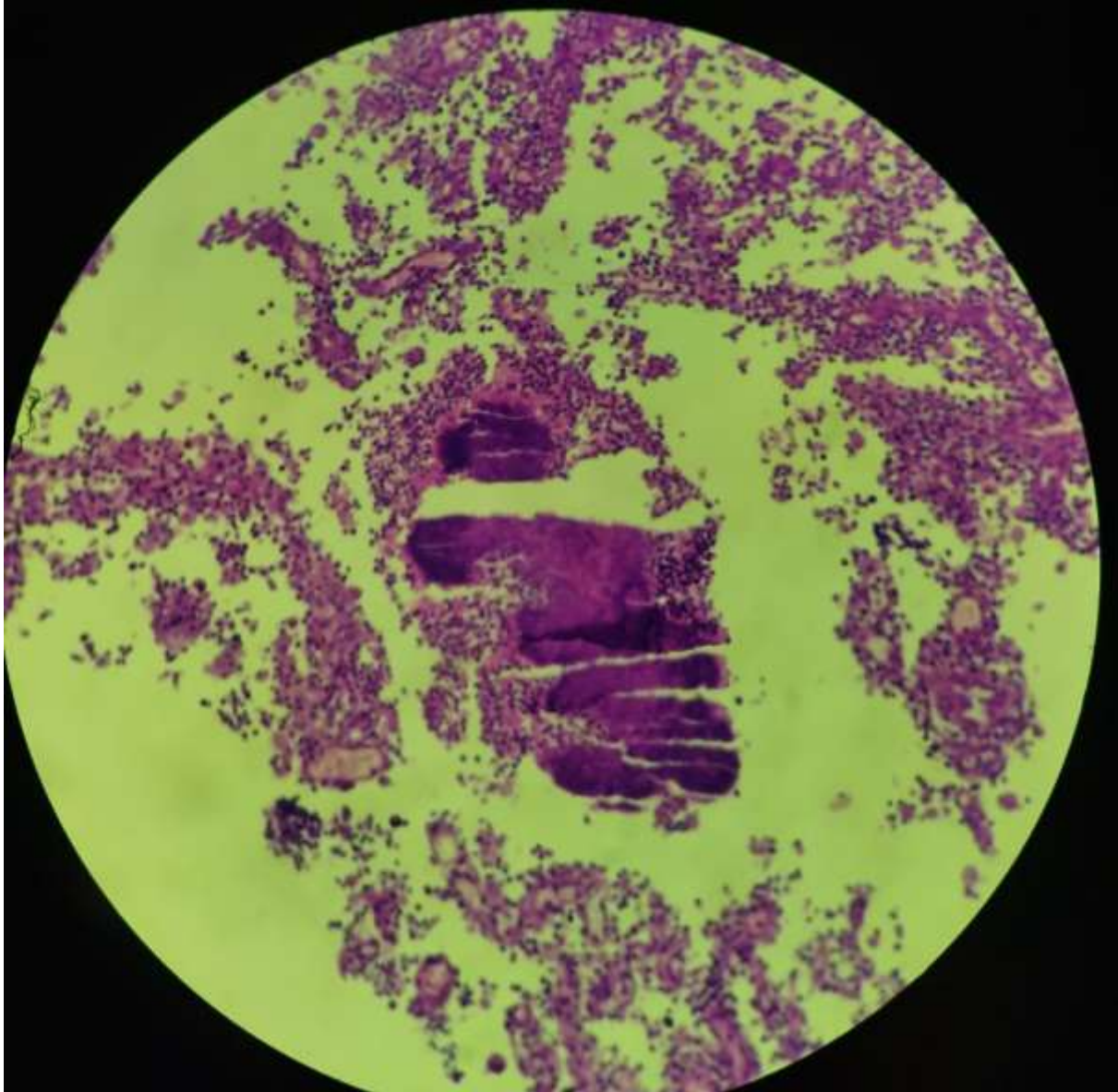


Figure 3:- Image of a microscopic examination (H&E stain x40) revealing the presence of actinomycetes sulfur granules surrounded by polymorphonuclear neutrophils.

Discussion:-

Actinomycosis is a filamentous bacteria discovered in 1880. It was long ago mistaken as fungi [2]. The usual human pathogen is *Actinomyces israeli*, which is a saprophytic anaerobic bacterium found in the oral cavity (teeth, tonsils), the digestive tract and the genital tract in women [3]. Contamination is often secondary to a breach of the mucosal barrier under trauma, surgery, or in the presence of other bacteria when associated with some illness such as diabetes or neoplasm [4], [5]. The exact etiology in our patient could not be determined. After the invasion of the bacteria, single or multiple abscesses will form and progressively evolve toward suppuration and fibrosis. The chronic course of the infection is the formation of a fistula that drains the lesion with the possibility of extension to some other organs, even joints and the central nervous system[5].

The most frequent bacteria in cause is: *Actinomyces israeli* (73%). However, there are over 30 species with variable morphological aspects and only 6 of them are harmful to humans. [6]

The bacteria predominantly affect men with a sex ratio of 3/1. It can occur at any age with an adult predominance and peak frequency at 40 years old [5]. Its incidence rate is hard to determine because diagnosis is often hard to establish. Mortality rate is estimated from 0-28%. [7]

Cervico-facial (60%), and thoracic (15%) locations are the most frequent [8], [1] As for the abdominal location (20-25%), actinomycosis frequently occurs in the ileocecal region in 67% of cases, including 32% in the appendix [4], [5], 15% in the colon and anorectal region, and 18% in the liver, biliary tract, pancreas, stomach, small bowel, and esophagus. [9]

The ileocecal localization often occurs after an interval of a few months, and such was the case for our patient (one month). It may also occur a few years after an appendectomy [5].

For abdominal localizations, actinomycosis can be revealed by clinical presentation of a localized peritonitis, or in other cases as an intra-abdominal abscess, an abdominal mass, or simulating Crohn's disease with inflammation of the ileocecal region and formation of fistula (which is the case for our patient).

Actinomycosis can affect the anus, the rectum, the colon, the caecum, the ileum, the endometrium, the liver, or the peritoneum. The infection can be triggered by a surgical procedure, a sigmoiditis, a diverticulosis, an appendicitis, a cholecystitis, an uncontrolled intrauterine device, or an abortion [5].

The diagnosis is often made during the postoperative course, and after an anatomopathological analysis of the surgical specimen. Actinomyces form filamentous aggregates whose ends are swollen with glycopolysaccharides (proteins and calcium phosphate), thus forming sulfur grains which do not contain sulfur. But they are named so for their yellow color. These sulfur grains have a diameter of less than one millimeter and are visible to the naked eye or under an optical microscope in which they appear like a net of fragile threads. The inflammatory granuloma that develops around the grains is formed of three concentric zones: a central area with altered leukocyte, a crown of histiocytic cells and an intense fibroblastic reaction and peripheral collagen that constitute an abscess which can become fistulized at any moment. [3], [6]

As for the clinical presentations, actinomycosis of the ileocecal region has no specific symptoms and it can take the form of acute appendicitis with right iliac fossa pain and fever and bowel disorders, such as diarrhea or constipation with a biological inflammatory syndrome. In its chronic form, it mimics Crohn's disease or a right iliac fossa mass.

Imaging is not accurate for the diagnosis of actinomycosis as it is often confused with an inflammatory or tumoral disease.

Bacteriological diagnosis is difficult because of the sensitivity of the germ to oxygen and the difficulty to perform a culture test. It is usually inconclusive if pathology is not suspected; however, if suspected, a prolonged bacteria culture test in an anaerobic environment can be highly suggestive of a diagnosis [1].

Endoscopy examination may show an infiltrative mass with yellow exudate [6].

Biopsies performed by endoscopy or surgical biopsies of infected site or pus are the best way to confirm the diagnosis. Many samples should be taken and accessed quickly to the lab for prolonged bacteria culture tests in an anaerobic environment. Negative results do not eliminate diagnosis.

Pathological findings may include the presence of characteristic microorganisms: some filamentous gram-positive bacteria with culture showing yellow sulfur granules with necrosis. [10]

Differential diagnosis can be made with tumors because of its insidious and infiltrative character. However, other infectious diseases such as tuberculosis or inflammatory disease such as Crohn can represent a main differential diagnosis especially with the presence of a granulomatous reaction in biopsies (tuberculosis) or an inflammation and a fistula (In the case of Crohn's disease).

The curative treatment is based on Beta Lactam antibiotics especially Penicillin G or Amoxicillin. [1]

Penicillin G is the treatment of choice at high doses of 3 to 5 million units intravenously every 6 hours, for 2 to 6 weeks. Afterwards, there is a need to switch to an oral antibiotic therapy with the dose of 1 gram every 6 hours. The use of ampicillin (6-12 g IV), cyclins, macrolides, or sulfonamides can be an alternative to penicillin. These bacteria are naturally resistant to aminoglycosides and metronidazole. The best treatment is an intravenous penicillin G followed by an oral amoxicillin (3-4 g per day). The duration of the treatment is up from 3 to 4 months after the disappearance of clinical symptoms for the digestive form, and from 6 to 12 months for the other forms [11]. Depending on the localization and the response to antibiotics, a shorter therapy may be considered [1]. This long-term treatment is necessary because of the poor diffusion of antibiotics into fibrosis.

Surgical treatment is indicated in acute forms: peritonitis, intestinal obstruction, or in case of doubt with a neoplastic disease. Percutaneous drainage can be used in case of abscess.

A clinical and radiological (ultrasound, CT) follow up is necessary to make sure the infection is resolving and for a follow up in patients with defects, such as diabetes or immunodeficiency, because there is a risk for some lesions to appear up to 10 years after the disease's onset. [12], [6], [10]

The prognosis of this infection is good if a well-adapted antibiotic therapy is initiated and if surgery is performed in the appropriate indication.

The evolution was favorable for our patient without any complications or recurrence during the first postoperative year.

Conclusion:-

Digestive actinomycosis diagnosis is very challenging and often confused with inflammatory diseases such as Crohn's disease, or infectious (appendicitis if acute or tuberculosis) or neoplastic pathology of the ileocecal region, often leading to unjustified or mutilating surgical procedures. Radiological diagnosis may be inconclusive. Only biopsies with anatomopathological analysis can confirm diagnosis.

Abbreviations:

1. CEA: CarcinoEmbronic Antigen
2. CRP: C Reactive Protein
3. CT: Computed Tomography
4. PT: Prothrombin Time
5. US: Ultrasonography

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