

Case Report

Isolated and fistulized primary appendicular tuberculosis: when a great pretender disguises herself

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ABSTRACT

Tuberculosis in all its forms remains a public health problem in Morocco, despite the efforts of the state in terms of prevention and treatment. Recently, there has been an increase in the number of extra-pulmonary forms of TB. Isolated appendicular localization is very rare, especially since intestinal localization is only ranked 6th among extra-pulmonary localizations. We report the case of a 39 year old female patient admitted for right iliac fossa (RIF) pain evolving for two months, the clinical examination on admission objective a digestive fluid outlet through a fistulous orifice at the level of the RIF; the abdominal CT scan found a plastron with the presence of a fistulous path between a digestive segment and the wall, the surgical exploration found a fistula between the body of the appendix and the wall, an appendectomy was done and the anatomopathological examination confirmed the tuberculosis origin. Our work sheds light on an often-misunderstood form of a well-known pathology.

Keywords: Tuberculosis, Appendicular, Enterocutaneous fistula

INTRODUCTION

Tuberculosis is an infectious disease caused by *Mycobacterium tuberculosis*, it is the 9th cause of death worldwide and the 1st cause in relation to a single infectious agent, even before the human immunodeficiency virus.¹ The intestinal localization is characterized by various digestive manifestations and can cause mainly in its ileocecal localization, a diagnostic problem mainly with Crohn's disease.² According to necropsy studies, the affected areas in increasing order are: duodenum, colon, ileocecal region and ileum.³ Isolated tuberculosis of the appendix is rarely reported.³

CASE REPORT

We report a rare case of appendicular TBK since it is associated with an enterocutaneous fistula. Our aim is to draw attention to this condition and to enrich the scientific literature on this grey area. We report the case

of a 39-year-old female patient, well vaccinated according to the Moroccan national immunization program (BCG in the first week of life), under psychotropic drugs for schizophrenia, admitted to the emergency room for IDF pain evolving for 2 months associated with slight asthenia, anorexia and weight loss judged minimal by the patient.

The clinical examination found a fistulous orifice located at the level of the IDF with the release of some fecal debris, the palpation found a sensitivity of the IDF associated with an impaction, the rest of the somatic examination was without particularities. The biological workup found hyperleukocytosis and an elevated CRP. The abdominal CT scan with injection of contrast medium was done and found: agglutination of some ileal loops in the right latero-uterine and adhesion with the right adnexa; evidence of an enterocutaneous fistula at the level of the IDF from the latero-uterine agglutinated loops (Figure 1).

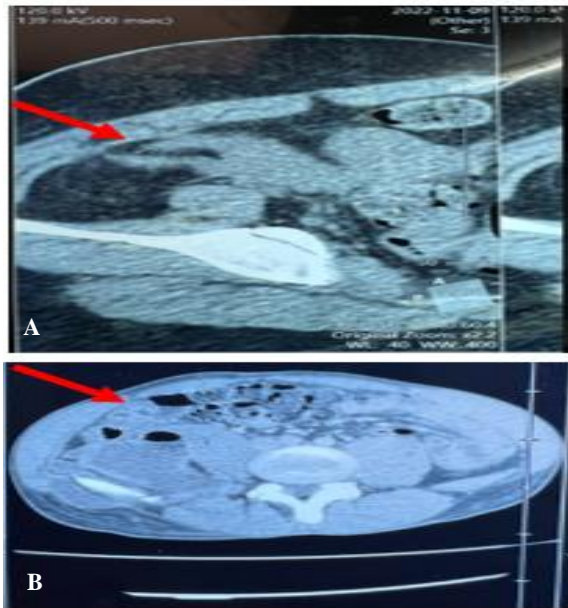


Figure 1 (A and B): Scannographic images showing the enterocutaneous fistula.

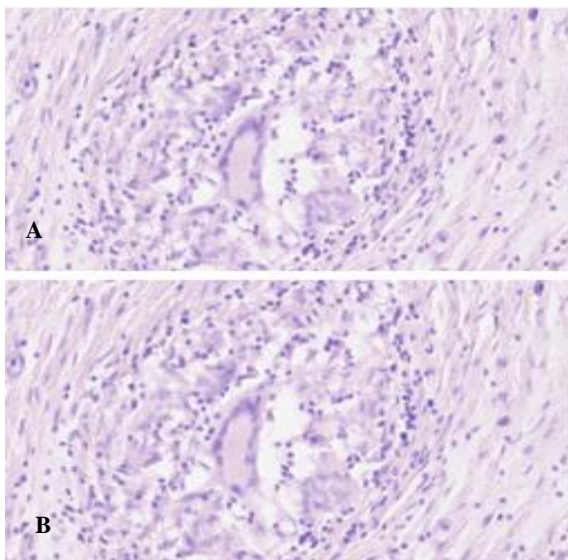


Figure 2 (A and B): Microscopic sections showing the epithelioid-giganto-cellular granuloma.

It was decided to intervene surgically by a median laparotomy under umbilical, the surgical exploration finds a plastron of the IDF fiat of the cecum, last ileal loop, great omentum, ovary and right tube around a thickened vermicular appendix with a healthy base and whose body communicates with the abdominal wall in front by a fistulous course. The surgical procedure consisted of release of adhesions, appendectomy, blinding of the fistulous path and peritoneal lavage. The postoperative course was unremarkable. The anatomopathologist answered that it was a granulomatous and tuberculoid pan-appendicitis with foci of caseous necrosis (Figure 2), the patient was entrusted to the infectious diseases department for further treatment.

DISCUSSION

Worldwide, tuberculosis is on the rise in all its forms, 1 in 5 patients with tuberculosis in the European Union has an extra-pulmonary form, intestinal tuberculosis ranks 6th among extra-pulmonary localizations and the appendix is incriminated in only 1.5 to 3% of all cases of intestinal tuberculosis.⁴⁻⁷ Theoretically, digestive tuberculosis can reach all segments with an increased tropism for parts rich in lymphoid tissue such as the terminal ileum and the ileo-caecal junction posing a serious diagnostic problem with Crohn's disease, the appendix is incriminated in only 1.5 to 3% of cases.⁶⁻⁸ The pathogenesis of intestinal tuberculosis, including appendicular location, is poorly understood. Three routes of contamination are possible: hematogenous contamination, local extension or from contaminated sputum swallowed by patients with active pulmonary tuberculosis.^{2,9} Appendicular tuberculosis has 3 anatomical forms: Chronic: characterized by a succession of chronically evolving waves of abdominal pain associated with vomiting and diarrhea, Latent: the appendix is macroscopically almost normal, the diagnosis is made by histology, Acute: the picture is that of a classic suppurated appendicitis.⁹

Macroscopically, appendicular tuberculosis can be divided into 3 main aspects: Normal, Hypertrophic, pseudotumor and Ulcerated: which may be complicated by abscess, perforation or fistula.⁹ Tuberculosis must be suspected at the slightest doubt, even if the history does not reveal any notion of contagion.¹⁰ It classically occurs in economically disadvantaged and unvaccinated subjects.¹⁰ The present case illustrates the diagnostic difficulty since it does not meet these socio-economic and demographic criteria. Clinically, appendicular tuberculosis is most often manifested by a classic appendicular syndrome to which is added weight loss (66%), fever (35-50%) and diarrhea (20%), which is evidence of tuberculosis impregnation.^{9,11} The diagnosis should be suspected and evoked whenever the abdominal symptoms are suspicious, the patient is living in an endemic area and is immunocompromised.¹ The paraclinical evaluation must consider the epidemiological situation of the country and the material resources available for this purpose.¹

The diagnostic approach remains problematic; the biology is not at all specific, with a biological inflammatory syndrome with elevated white blood cells, anemia and increased CRP.^{12,13} The intradermal reaction is neither sensitive nor specific.⁹ The interest of the QuantiFERON-TB test lies essentially in the absence of cross-reaction with the BCG vaccine, which may be responsible for an intradermal reaction > 5 mm in the absence of tuberculosis. Thus, the QuantiFERON-TB is more specific than the intradermal reaction in the BCG-vaccinated population.² Intraoperative diagnosis is almost impossible; The histological study brings the diagnosis of certainty by the demonstration of an epithelioid and

gigantocellular granuloma centered by a caseous necrosis and delimited by a lymphocytic corona, it is noted that the distinction between Crohn's disease and tuberculosis is hardly easy, especially since their coexistence is possible at least theoretically, especially during biological treatment in immunocompromised persons, exudative and necrotizing forms are more frequent, thus requiring Ziehl's staining for the detection of the Koch's bacillus, thus allowing the elimination of a certain number of differential diagnoses: actinomycosis, histoplasmosis, yersiniosis or Crohn's disease which is only possible in 26 to 36% of cases.^{1,2,9,14}

The treatment of appendicular tuberculosis is multimodal; surgery should only be performed in the event of complications; a simple appendectomy has been proposed by some authors as a treatment for isolated appendicular tuberculosis; this attitude has been abandoned and antibiologic chemotherapy should always be administered for 6 months based on (Streptomycin, Rifampicin, Isoniazid, Pyrazinamide) for two months and Rifampicin, Isoniazid for four months.^{1,15} In case of persistent diagnostic doubts, a trial antibiologic treatment must be initiated and only the response to this treatment is taken as a major diagnostic criterion.¹⁶ The parenteral route is preferred in case of incoercible vomiting or digestive malabsorption; treatment should be continued for up to 9 months in patients with AIDS.⁴ Resistance to chemotherapy is possible, a situation that requires rapid and appropriate action.¹⁷

CONCLUSION

Appendicular tuberculosis is a rare epidemic-clinical-histological entity. Its pathophysiology remains poorly elucidated. Clinically, appendicular tuberculosis in its isolated form simulates the picture of acute appendicitis. Diagnostic confirmation is histological although it is not always easy. The addition of antibiologic drugs to surgical treatment is accepted as the standard of care.

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