

## Case Report

# Brucella terminal ileitis: a rare cause of intestinal obstruction about a case

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## ABSTRACT

Brucellosis is a bacterial zoonosis transmitted by contact with fluids from infected domestic animals, by consumption of unpasteurized milk products, or by inhalation of infected aerosols. Systemic infection has clinical manifestations from asymptomatic cases to those that are fatal. Focal infections occur in 30% of cases and affect any organ. Gastrointestinal manifestations are rare and unspecific. We reported the case of a patient with antithrombin III deficiency who presented with a clinical picture of abdominal pain at the emergency department, with failure of conservative treatment and with progression to acute abdomen, for which surgical treatment was offered, resolving the condition of intestinal obstruction and taking a biopsy that confirmed terminal brucella ileitis, antibiotic treatment and intestinal rest were indicated with successful results. Brucella terminal ileitis is an unusual manifestation of brucellosis. History of consumption of unpasteurized milk and derivatives and contact with livestock should be carefully examined in patients with acute abdomen in Brucella endemic countries. This will lead to a full and uncomplicated recovery from this disease.

**Keywords:** Intestinal obstruction, Brucellosis, Acute abdomen, Zoonosis, Terminal ileitis, Antithrombin deficiency

## INTRODUCTION

Brucellosis is a bacterial zoonosis transmitted by contact with fluids from infected animals (sheep, cows, goats, pigs), by consumption of unpasteurized milk products or by inhalation of infected aerosols. There are several species of Brucella: *B. melitensis* (small ruminants), *B. abortus* (cattle), *B. suis* (pigs) and *B. canis* (dogs) that cause disease in humans.<sup>1</sup> The origin of human brucellosis is lost in history, although the first clinical report is attributed to Marston in 1859. The etiological agent was discovered at the end of the 19th century by Sir David Bruce, who was sent to investigate on the Island of Malta. The cause of a feverish illness that had caused the death of a considerable number of soldiers. The germ was identified in 1887 in the spleens of four deceased soldiers and was named *Micrococcus melite*.

In 1896 Bang, a Danish veterinarian discovered the causative agent of bovine abortion, which in the future was called *Brucella abortus* and in 1905 Themistokles Zammit documented the role of goats and the consumption of their products, as a source of contagion to acquire the disease.<sup>2</sup>

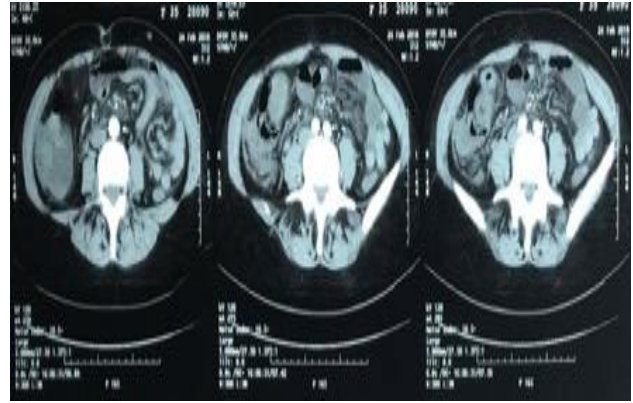
The incidence figures for human brucellosis in Mexico are imprecise, due to underreporting; however, it is reported as the second place in America, with an incidence of 2.97 cases per 100,000 inhabitants in 2011. Mortality is less than 1% with adequate antibiotic treatment.<sup>1</sup>

It is a systemic infection with versatile presentations and systemic complications, however, gastrointestinal complications are very rare.<sup>7</sup> This report presents a rare case of a woman who presented terminal ileitis and secondary intestinal obstruction due to Brucella infection.

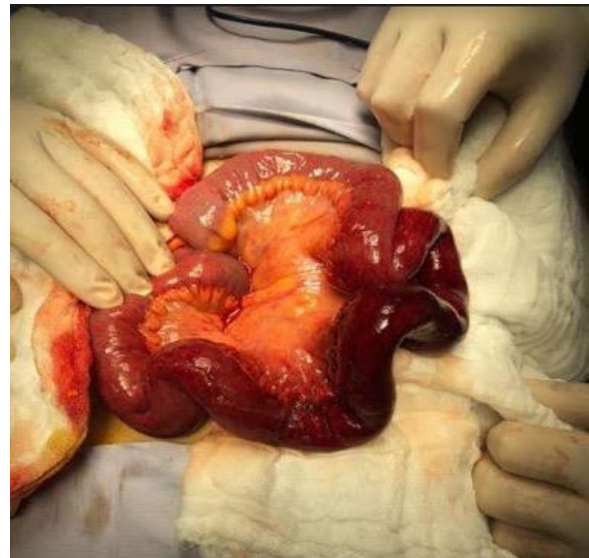
## CASE REPORT

The case of a 56-years-old female patient with antithrombin III deficiency under treatment with rivaroxaban 20 mg per day and a history of allergy to tetracyclines is presented. His condition began 12 hours prior to his hospital admission with sudden abdominal pain, located in the lower hemiabdomen, in intensity 10/10 on a numerical scale, accompanied by nausea and vomiting. On physical examination with data from the acute abdomen. It is managed with analgesics, crystalloid solutions and paraclinical tests are requested, radiographs of the abdomen in decubitus and standing are observed fixed loop in the right iliac fossa (Figure 1), hematic biometry with leukocytosis and neutrophilia with bandemia, simple and contrasted tomography of abdomen there is dilation of the small intestine loops, thickening of the terminal ileum wall and abdominal free fluid (Figure 2), an exploratory laparotomy is performed, with findings of intestinal obstruction secondary to adhesions at the terminal ileum and ischemia in a segment of 30 cm to 5 cm of ileocecal valve and mesenteric lymphadenitis.

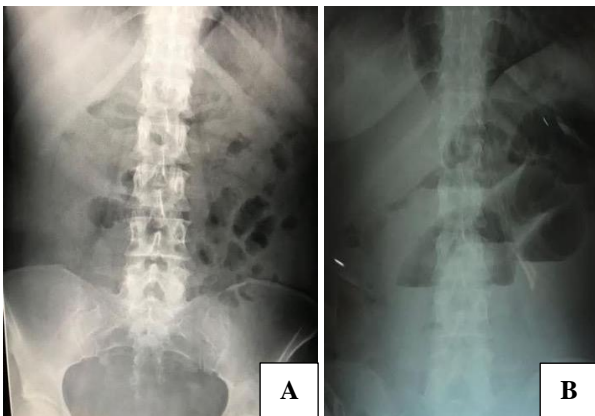
Adherolysis is performed, reversing the ischemia data, and thickening of all the layers of the terminal ileum is palpated without perforations or data of venous or arterial thrombosis in the mesentery. After the affected 30 cm, the rest of the normal digestive tract is found. do not perform intestinal resection and biopsies of the lymph node and mesentery are taken and the surgical procedure is completed (Figure 3). Infectious pathology of terminal ileum versus Crohn's disease is suspected, a Rose Bengal test is taken, reactive 1: 100, brucellosis is confirmed with a 2-mercaptoethanol agglutination test, positive 1:80 for *Brucella abortus* (Figure 4). Treatment with rifampicin and trimethoprim with sulfamethoxazole was started due to a history of allergy to doxycycline. The evolution was favorable after the intervention with analgesic treatment, antibiotic therapy and intestinal rest. After days of remission of the clinical picture, without the presence of complications, the patient was discharged and sent to the general surgery outpatient clinic to continue monitoring.



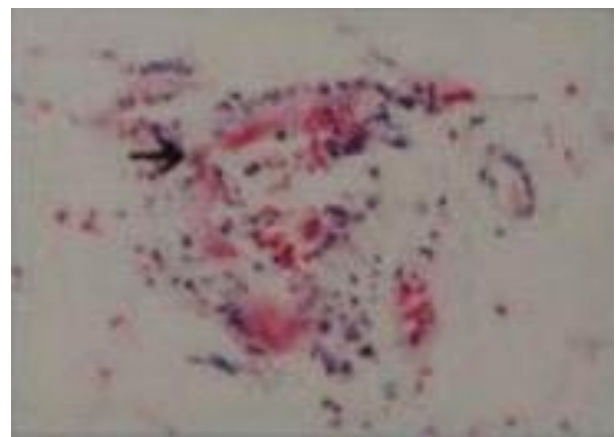
**Figure 2: Simple and contrasted tomography of the abdomen in axial sections showing dilation of the small intestine loops, thickening of the terminal ileum wall and free intra-abdominal fluid.**



**Figure 3: Intra-operative image showing ischemia at the level of the terminal ileum in a segment of 30 cm to 5 cm of the ileocecal valve and mesenteric lymphadenitis.**



**Figure 1: Plain X-ray of the abdomen in standing and decubitus projection showing a fixed loop in the right iliac fossa and air-fluid levels.**



**Figure 4: Representative sample of mesenteric ganglion HE staining obtained from the patient.**

## DISCUSSION

Brucellosis, also known as ‘undulating fever’, is produced by a gram-negative coccobacillary bacteria, 0.5 to 0.7  $\mu\text{m}$  in diameter by 0.6-1.5  $\mu\text{m}$  long, intracellular, facultative, non-sporulated, without capsule, Brucella species have a capacity unique to invade phagocytic and non-phagocytic cells, and ability to survive in an intracellular environment by finding ways to bypass the immune system. Its cytoplasm is rich in DNA, RNA and cytosolic proteins, some of them important for its diagnosis.<sup>3</sup>

The disease is acquired by different routes such as direct contact with animals, their excreta, or from the environment where Brucella has survived, orally, parenterally, nasal or inhalation. Person-to-person transmission is rare but can occur. By the oral route (anthropozoonosis) the bacteria pass to the digestive system, the main source of this infection route being the ingestion of unpasteurized milk or dairy products, especially those from sheep and goats without ruling out those from cattle.<sup>3</sup>

In humans, Brucellosis can be produced by *B. abortus*, *B. melitensis*, *B. suis* biotypes 1-4 and sometimes by *B. canis* or Brucella of marine mammals, *B. ovis*, *B. neotomae* are also pathogenic to humans.<sup>4</sup>

Incubation ranges from one to four weeks to several months. Systemic infection has clinical manifestations ranging from asymptomatic to fatal complications. Focal infections occur in 30% of cases and affect any organ: osteoarticular, genitourinary, pulmonary, hematological, gastrointestinal, neurological, cardiac, ocular or dermatological.<sup>1</sup>

Gastrointestinal manifestations are diverse and are often characterized by loss of appetite, vomiting, and abdominal pain. Colitis and ileitis have been reported as rare sporadic cases in the literature.<sup>4</sup> In a meta-analysis of the neglected tropical diseases that included 57 articles related to the most frequent manifestations of brucellosis and that included 12 countries, it concluded that the abdominal manifestations were pain, splenomegaly and hepatomegaly with estimated overall proportions of 19%, 26% and 23% respectively. The number of studies that reported the presence of hepatitis was small, only seven in total, with an estimated 4% of patients affected in general.<sup>5</sup> Similarly, another meta-analysis carried out in China established hepatitis as the most frequent abdominal complication, not including ileitis or intestinal obstruction at any intestinal level in their results.<sup>6</sup> The infection can present as an acute abdomen that simulates acute appendicitis, ileitis, infectious colitis, acute cholecystitis, acute pancreatitis, mesenteric lymphadenitis and even intestinal occlusion as in this case report.<sup>7</sup>

The clinical diagnosis of brucellosis can be difficult due to its nonspecific signs and symptoms, so a high index of suspicion can lead to the correct diagnosis. It is important

to obtain a detailed history that includes recent exposure to common Brucella host species, especially cattle, sheep, goats, pigs, camels, buffalo, or dogs; consumption of raw or undercooked milk or dairy products, meat and offal derived from these animals.

Occupational exposure, travel, or residence in an area where infection is prevalent also increases the likelihood of diagnosis. The diagnosis is made with certainty when brucella is isolated from blood, bone marrow, or other tissues. Serological tests are the most widely used method to diagnose brucellosis, among which the standard agglutination test is considered positive at titers of 1/160 or more in case of active infection.<sup>8</sup> The ELISA for diagnosis of Brucella is the most common serological test. sensitive and specific, and can be positive when other tests do not. Recently, the polymerase chain reaction (PCR) has been developed for the detection of Brucella species in human blood samples. The PCR test for samples of other secretions has also been described. Different tests can be combined to improve diagnostic performance.<sup>9</sup> A combination of PCR+ELISA tests appears to be a highly sensitive and specific diagnostic method.<sup>8</sup>

In Mexico there are multiple antimicrobial regimens for the treatment against Brucella. These regimens include the mixture of two antimicrobials of choice, among them are tetracyclines (doxycycline), aminoglycosides (streptomycin), rifamycin (rifampicin), trimethoprim/sulfamethoxazole and fluoroquinolones (ciprofloxacin and levofloxacin). Due to the history of allergy to the tetracyclines group in our case report, the treatment of choice for the patient was rifampicin+trimethoprim/sulfamethoxazole indicated for 21 days.<sup>10</sup>

## CONCLUSION

Brucella terminal ileitis is an unusual manifestation of brucellosis. History of consumption of unpasteurized milk and derivatives and contact with livestock should be carefully examined in patients with acute abdomen in Brucella endemic countries. This will lead to a full and uncomplicated recovery from this disease. Despite a history of allergy to a first-line drug for the treatment of this infection, the patient presented in our clinical case was successfully treated, avoiding complications that put her life at risk.

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