LETTER TO THE EDITOR

Splenic abscess as the first manifestation of Crohn’s disease

Dear Sir,

This report describes a case of Crohn’s disease presented with a rare extraintestinal complication: splenic abscess.

A 60-year-old male without antecedents of interest, consulted for fever of 38–39 °C mainly in the evening for the past month, accompanied by shivering, profuse sweating, left flank pain irradiated to the shoulder and weight loss of 4 kg. He referred placement of dental implants two months before onset of symptoms.

On examination, the patient was febrile, with altered general state and painful splenomegaly. Blood test showed leukocytosis (16,400/L, 86.1% neutrophils), haemoglobin 117 g/L, ESR 68 mm in the 1st hour, pattern of dissociated cholestasis (AST 19U/L, ALT 26U/L, ALP 203U/L, GGT 213U/L, total Bb 0.5 mg/dL). No alterations were observed in urine sediment and thoracic X-ray was normal.

Abdominal ultrasonography revealed splenomegaly and presence of multiple anechoic hypoechogenic nodules suggestive of splenic abscesses (Fig. 1). Abdominal CT confirmed the presence of abscesses and detected small retroperitoneal adenopathies, with no other findings. Treatment with ceftriaxone and metronidazole was started and puncture aspiration of one of the splenic nodules was positive for Streptococcus mitis. Haemocultures were negative. Transoesophageal echocardiography ruled out infective endocarditis.

As fever persisted despite antibiotic treatment and general status of patient worsened, abdominal CT was repeated, which showed progression of splenic abscesses and splenectomy was programmed. Anatomopathological study confirmed multiple splenic abscesses. PAS/PAS diastase, Gomori, Giemsa, Kinyoun and Mucicarmin stains did not detect microorganisms, lymphoproliferative process or histopathological signs suggestive of malignancy. The patient was discharged and treatment with ceftriaxone was maintained for 6 weeks.

One month later, the patient was readmitted for recurrence of fever, abdominal pain, occasional vomits, progressive weight loss and diarrhoea. Blood cultures and coprocultures (including culture and detection of Clostridium difficile) were repeatedly negative. Abdominal CT ruled out post-surgical complications and showed duodenal wall thickening and mesenteric adenopathies. Enteroscopy revealed the presence of multiple lineal and circular ulcers in the 3rd and 4th sections of duodenum and proximal jejunum. Anatomopathological study showed superficial ulceration of mucosa and signs of moderately severe acute and chronic inflammations in lamina propria without granulomas. Immunohistochemical staining was negative for CMV. Fibrocolonoscopy detected small erosion in the cecum with normal terminal ileum.

The study was completed with determinations of immunoglobulins, thyroid hormones, tumour markers, immunological study for antibodies, screening for celiac disease, hormonal study, viral and bacterial serologies (HBV, HCV, CMV, Leishmania, HIV, Cryptococcus, Toxoplasma, Brucella, Yersinia, Bartonella, Francisella tularensi, and Coxiella) and basic study of humoral and cellular immunity – all within normal limits. As symptoms persisted and patient’s general state worsened, enteroscopy was repeated and samples were collected for microbiological study (PCR mycobacterium, T. whipplei and Bartonella negative). Histology revealed distortion of crypt architecture, substantially increased inflammatory cellularity in the lamina propria, cryptitis and presence of occasional epithelioid granuloma with negative PAS and PAS diastase stain; all highly suggestive of an inflammatory bowel disease such as Crohn.

Corticosteroid therapy was initiated at a dose of 1 mg/kg, with clear improvement, the fever disappeared and diarrhoea remitted completely.
Treatment was subsequently changed to concomitant endovenous infliximab and azathioprine; because the patient had hyperosmolar non-ketotic decompensation with corticosteroid treatment. Doses were administered every 6 weeks for exacerbation of symptoms.

Extraintestinal manifestations of Crohn's disease are well documented. These involvements may precede, occur with, or follow exacerbations of inflammatory bowel disease. Splenic abscess is very uncommon because the spleen is an effective filter for organisms. The causes of splenic abscesses often fall into five categories: metastatic infection, superinfection following ischaemia or infarction due to red blood cell abnormalities, trauma, contiguous infection and immunodeficiency.1

The clinical presentation of splenic abscess is insidious, often with constitutional symptoms: fever, nausea, general malaise and abdominal pain localized in left upper quadrant.

The nonspecific nature of the signs and symptoms of splenic abscess makes imaging studies a cornerstone of diagnosis. CT has a sensitivity of 96% and is superior to US, which has a sensitivity of 76–93%. US allows percutaneous puncture and CT is superior in its ability to localize small lesions and provides better information about the perisplenic area and contiguous viscus.2

Blood cultures were positive in 24-60% of cases,3 whereas the culture of the material obtained from the abscess was positive in >90%. In our patient, the blood cultures were repeatedly negative, probably because he had received various antibiotic treatments. Streptococci, Staphylococci, Salmonellae and Escherichia coli are usually the major bacterial causative agents of splenic abscesses. Anaerobic bacteria remain a relatively infrequent aetiology (5–7%). However, the increasing number of immunocompromised patients reflects a greater number of fungal and mycobacterial infections. It should be stressed that the culture is sterile in up to 25% of the cases.4 In our case, the microorganism isolated by aspiration puncture of one of the splenic lesions was S. mitis, a Streptococcus viridans that frequently colonizes the oral cavity, respiratory and gastrointestinal flora and is responsible for 6–8% of the episodes of bacteraemia in our country.5

Treatment of splenic abscess is controversial. Splenectomy has been the treatment of choice to date, but increasing experience and treatment success have been reported with percutaneous puncture and drainage. However, contraindications for this modality are multilocular abscesses, thick septations and abscess rupture with bleeding.3

Systemic involvement in inflammatory bowel disease is well known. Extraintestinal symptoms occur in 30–40% of patients with Crohn's disease and are the initial symptoms in 10% of cases.

We conducted a literature search covering the past 30 years using the MEDLINE database, and only found two reports4,7 where splenic abscess was identified as the initial manifestation of Crohn's disease. The first refers a patient who presented other signs and symptoms for initially suspecting the diagnosis of inflammatory bowel disease.6 The other report refers seven cases of sterile abscesses with a granulomatous pattern on histopathological examination (single splenic abscess (1 case), spleen and other locations (4 cases), no splenic abscess (1 case)) that preceded the diagnosis of Crohn's disease with a range from 1 to 41 months.7 In the other articles reviewed, splenic abscesses appear in patients already diagnosed of Crohn's disease with contingency of bowel disease or colon-splenic fistula. Abscesses are often secondary to injury of ulcer perforation or a fissure, or secondary to transmural inflammation.8 The intraabdominal location (perineal, perirectal or associated with enterosenteral fistulas) of these abscesses is relatively common (15–30%), unlike the splenic, retroperitoneal and liver locations, which are quite rare.9

The diagnosis of Crohn's disease is usually established with endoscopic findings in a patient with a compatible clinical history and the intestinal biopsy is usually confirmatory rather than diagnostic. The major findings are focal ulcers and acute chronic inflammation. Granulomas may be found in up to 30% of patients with Crohn's disease and are diagnostic of the disorder if appropriate infections are excluded. In our case, the atypical presentation of the disease which seemed suggestive of an infectious disease and the unspecificity of the first intestinal biopsy delayed the definitive diagnosis until a second biopsy demonstrated the presence of cryptitis and granulomas.

References


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