CLINICAL CASE

Spontaneous splenic rupture, an unusual presentation of tuberculosis

Ruptura esplénica espontánea, una presentación inusual de tuberculosis

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Abstract

Introduction: *Spontaneous splenic rupture from tuberculosis (TB) is a very unusual presentation within the wide range of presentations of this infectious disease.* **Clinical case: A** *40-year-old male with a diagnosis of human immunodeficiency virus, begins with fever and pain in the left hypochondrium. A computed tomography scan was performed, showing probable splenic abscesses; suddenly, it begins with hemodynamic deterioration, exacerbation of pain, a surgical exploration was performed, showing spontaneous splenic rupture. Microscopic study of the spleen shows the presence of Mycobacterium tuberculosis.* **Conclusions:** *This is yet another presentation of TB, which can become a surgical emergency.*

Key words: *Tuberculosis. Extrapulmonary. Rupture. Spleen. Spontaneous.*

Resumen

Introducción: *La ruptura esplénica espontánea por tuberculosis es una presentación muy inusual dentro de la amplia gama de presentaciones de esta enfermedad infectocontagiosa.* **Caso clínico:** *Masculino de 40 años con diagnóstico de VIH, inicia con fiebre y dolor en hipocondrio izquierdo. Se realiza TAC evidenciando probables abscesos esplénicos; súbitamente comienza con deterioro hemodinámico, agudización del dolor, se realiza exploración quirúrgica evidenciando ruptura esplénica espontánea. Al estudio microscópico del bazo se observa presencia de Mycobacterium Tuberculosis.* **Conclusiones:** *Esta es una presentación más de la TB, la cual puede convertirse en una urgencia quirúrgica.*

Palabras clave: *Tuberculosis. Extrapulmonar. Ruptura. Bazo. Espontáneo.*

Introduction

Tuberculosis (TB) continues to be a mayor health issue around the world despite advances in its diagnosis and treatment. This disease presents with an important clinical diversity, a characteristic that makes TB a challenge for the treating physician, whether clinical or surgical. Within this diversity, we can classify its presentation in pulmonary and extrapulmonary TB, the latter being the one that covers only 15% of all diagnosis[1].

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Extrapulmonary TB with abdominal origin is a rare condition in western countries, despite this, a resurgence of the disease has been observed in recent years in developed countries due to the increase in the frequency of immunodeficiency state cause mostly by human immunodeficiency virus (HIV)[1]. In spite of the fact that the presence of pulmonary TB can be associated with abdominal TB, only 15% of patients with abdominal TB have accurate evidence of pulmonary TB[2], this is relevant because it is these cases that become a greater challenge for its diagnosis and management. Of the organs and tissues involved in abdominal TB, splenic involvement has been reported in cases of disseminated TB, where immunosuppression is significant[3], while its isolated involvement remains an unusual entity[3-5]. Risk factors for splenic TB include immunosuppression, previous pyogenic splenic infections, a history of splenic trauma[6].

We presented a case of a male with fever of origin to determine to whom the presence of probable splenic abscesses is radiographically evidenced, later the patient suffers sudden clinical deterioration due to spontaneous splenic rupture.

Clinical case

We present the case of a 40-year-old male with no relevant history, who went to the ER for reporting fever of 2 months of evolution, intermittent, predominantly daytime, with maximum peaks recorded of 40°C, accompanied by asthenia, adynamia, loss of unintentional weight of about 5 kg in the past 2 months, and denied any other history.

During the initial evaluation, the patient was found to be feverish, tachycardia, with normal blood pressure, without integument pallor, or jaundice. On physical examination, the lung fields were auscultated without consolidating any pleuro-pulmonary syndrome, normal heart sounds without the presence of murmurs; on abdominal palpation, he found slight pain in the left hypochondrium, with no evidence of peritoneal irritation, in addition to the presence of Grade 1 splenomegaly without hepatomegaly, the rest of the examination without relevant data.

Within laboratory tests, the initial blood count showed a hemoglobin at 12.1 g/dL, leukocytes at 6.32 k/uL, neutrophils at 5.70 k/uL, lymphocytes at 0.448 k/uL, platelets at 147 k/uL, a preserved function of the kidney with serum creatinine of 0.8 mg/dL, and a slight increase in liver enzymes: aspartate aminotransferase 145 IU/L, alanine aminotransferase 99 IU/L, alkaline phosphatase 139 IU/L, in addition to total bilirubin 1.6 mg/dL, direct bilirubin 0.5 mg/dL, indirect at 1.1 mg/dL, and general urine test within normal limits. His initial chest and abdominal radiographs were observed without relevant data.

Viral serology was requested by immunoassay, being positive for HIV, so GeneXpert was performed detecting HIV-1 with a viral load of 3,260,000 copies/mL, with an absolute CD4 count of 66 c/μL. Samples were taken for blood culture which was negative for bacteria and fungi, including Ziehl-Neelsen and potassium hydroxide tests on sputum, intradermal reaction test for tuberculin (Purified Protein Derivative), coccidioidin and candidin, Venereal Disease Research Laboratory in blood and cerebrospinal fluid, which were negative.

His study was complemented with a computed axial tomography (CT) of the chest and contrast abdomen where the lung parenchyma was observed without the presence of infiltrates or pleural effusion, without cavitated lesions or the presence of mediastinal lymph nodes; in the abdomen, data of hepatic steatosis and splenomegaly of 13.6 cm were observed, associated with multiple hypodense images of diffuse distribution, the largest being 2.2 cm in diameter, suggesting multiple splenic abscesses (Fig. 1). Due to the presence of these images and the persistence of the clinical picture, it was planned to perform a splenectomy by laparoscopy, however, during the pre-operative period the patient presented sudden deterioration in his vital signs, required supplemental oxygen, tachycardia at 140 bpm, and hypotensive with 80/60 mmHg, which led to hemodynamic resuscitation based on crystalloids and blood products, which stabilized the patient. On exploring the abdomen, distention and stiffness were found, with evidence of frank peritoneal irritation. Given this scenario, it was decided to urgently enter the operating room; the left subcostal approach was started finding 500 cc of hemoperitoneum located in the left upper quadrant, the spleen was observed with the presence of multiple pinpoint whitish lesions in its parenchyma, in addition to the presence of active bleeding secondary to a Grade V injury. Vascular control of the splenic hilum was performed to perform the splenectomy, the rest of the abdominal cavity was normal. In figure 2, we can see the surgical piece resulting from the splenectomy.

During his immediate postoperative period, he continued to require vasopressors, so he went to the intensive care unit to continue his recovery. He

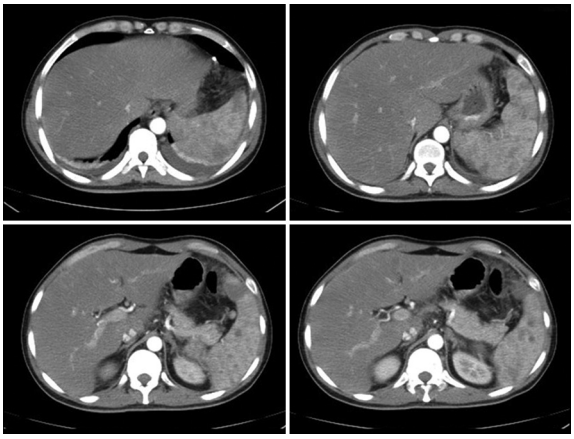

Figure 1. *Computed tomography of the abdomen where multiple hypodense lesions with diffuse distribution are observed in the splenic parenchyma, suggestive of splenic abscesses.*

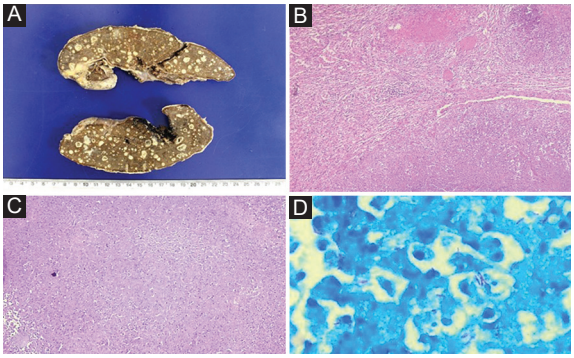

Figure 3. *At multiple sections grade V splenic rupture is evident, in addition to multiple well-circumscribed lesions.* **A:** *whitish brown in color and soft in consistency.* **B:** *on light microscopy, multiple granulomas.* **C:** *with a necrotic center were evident.* **D:** *Ziehl-Neelsen staining was positive for multiple acid-resistant bacilli.*

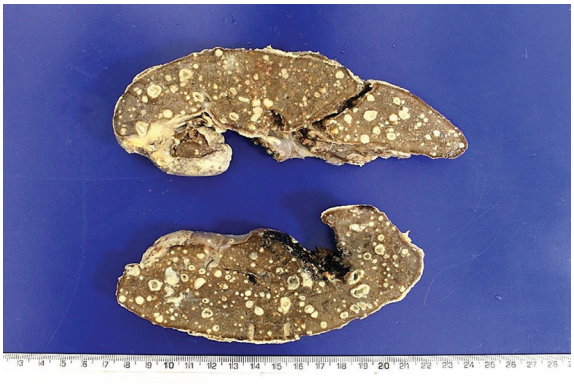

Figure 2. *Product of splenectomy, we observe the rupture of the splenic parenchyma in addition to the whitish nodular lesions throughout the spleen.*

presented hemodynamic and clinical improvement to the 10th post-operative day, so he continued his in-hospital management at the general hospital ward.

The histopathological report of the spleen reported a chronic granulomatous inflammatory process associated with the presence of acid-resistant bacilli (Fig. 3). GeneXpert was performed for *Mycobacterium tuberculosis* with a positive result and no resistance detected for rifampicin, so antifimic therapy based on rifampicin 150 mg, isoniazid 75 mg, pyrazinamide 400 mg, and ethambutol 300 mg in its intensive phase was started, responding favorably to treatment. Finally, and after 62 days of hospitalization, he was discharged with an outpatient follow-up.

Discussion

At present, it is estimated that 10 million people in the world (a range of 9-11 million) have been diagnosed with TB in 2018 alone, a number that has remained stable in recent years with a rate of 500 new cases for each 100,000 inhabitants per year; 251,000 deaths from HIV-positive TB are reported in these cases[7]. In Mexico, TB is an important public health problem since the binomial TB-HIV is a common presentation in our population. Due to this association of diseases, the clinical presentations can be so varied that they become exceptional in different cases.

Clinically, TB presents as a pulmonary or extra pulmonary disease, the most frequent being the first presentation[8]. Splenic TB is a singular and unusual case and can present itself in two ways: the first form is during miliary TB in immunocompromised patients, the spleen being the third most affected organ in miliary TB (100% lung, 82% liver, spleen 75%, lymph nodes 55%, and bone marrow 41%)[3-9]. The second unusual way of presenting is primarily, as it was in our patient, with few reports in the past 20 years[10]. These cases were immunocompromised patients who started their symptoms with fever of origin to be determined, which is characteristic in the presentation of splenic TB[11].

Primary splenic TB is presented as a case of hypersplenism, splenic abscess, or as a solitary splenic lesion. Its detected by contrasted CT that shows multiple hypodense nodulations in the splenic parenchyma; however, these can be characteristic of several conditions in addition to splenic TB such as cysts,

hematomas, and fungal infections such as candidiasis, spleen infarctions, lymphoma, or metastasis[12-14].

The diagnosis of splenic TB is complicated in patients without evidence of pulmonary involvement. Histopathological confirmation is required and this can be obtained in the first instance by means of a splenic biopsy, having an 88% sensitivity with fine-needle aspiration for the diagnosis of splenic TB[15]. The decision to perform a biopsy or go directly to splenectomy is entirely up to the doctor, depending on the conditions in which the patient is.

The histological findings of splenic TB are the same as those observed in the pulmonary presentation: the presence of granulomas with a necrotic center, surrounded by epithelioid histiocytes and a lymphocyte crown, either isolated or diffuse, together with the presence of bacilli acid-alcohol resistant in Ziehl–Neelsen staining. The presence of giant Langhans-type cells is variable, and is not exclusive to this disease. Likewise, other infectious possibilities should be ruled out with this histological image, especially *Coccidioides* Spp infections in our setting.

The treatment described in the literature is based on anti-TB drugs without actually performing a splenectomy[8], with them an adequate response is obtained at 6 months, similar to extrapulmonary TB from other sites due to the excellent penetration into tissues of this type of drugs. There are controlled clinical studies that recommend a duration of up to 12 months with the possible prolongation of treatment if necessary[16]. Due to the good response of the symptoms with anti-TB drugs, these remain the first line of the treatment for splenic TB and splenectomy is rarely required; however, surgical treatment is necessary if there is the formation of a splenic abscess, if the biopsy specimens do not show diagnostic results, when there is no response to treatment or when the patient's clinical conditions become critical, as it was in our patient[17].

Conclusions

The spectrum of presentations of TB is very broad, and primary splenic TB is itself a very rare medical entity in an immunocompromised patient. The spontaneous rupture adds a greater singularity to the case due to the low frequency with which it is reported. In this case, a probable spontaneous splenic rupture was suspected clinically due to sudden clinical deterioration, which was confirmed in the operating room, this being a highly unusual surgical emergency reported in the literature. It is important to consider the extent of TB, since it is an entity that can start its management with medical treatment and close monitoring; however, conditions can become an emergency situation where surgical treatment will be the difference between life and death in this type of patient.

Conflicts of interest

The authors declare no conflicts of interest.

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Ethical disclosures

Protection of human and animal subjects. The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

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