Case report

Spontaneous splenic rupture during infection of cytomegalovirus. A case report∗

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Abstract

Spleenic rupture is most commonly encountered after blunt abdominal trauma. Spontaneous atraumatic splenic rupture is a rare but dramatic occurrence that is most commonly attributed to infection or neoplasia. We report the case of a 27-year-old female patient without pathological history. Admitted to the emergency department for the sudden onset of left hypochondrial pain associated with vomiting, rapidly progressing to hypovolemic shock. She had reported an influenza-like illness a week earlier for which her COVID-19 PCR was negative. Emergency abdominal ultrasound and CT-scan revealed a ruptured spleen and widespread hemorrhagic fluid in the abdomen. Exploration revealed multiple ruptures in the spleen capsule. The patient underwent splenectomy with good clinical evolution. Despite the rarity of this condition, physicians should consider the diagnosis of spontaneous non traumatic splenic rupture when encountering healthy patients presenting with nonspecific left hypochondrial abdominal pain and hypovolemia. Mortality is essentially related to the delay in diagnosis and treatment and to the severity of the underlying pathology. Treatment often consists of splenectomy.

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Introduction

The spleen is an immunological organ affected by hematological and non-hematological diseases. Spleen rupture usually occurs because of blunt abdominal trauma. In contrast to traumatic spleen ruptures, spontaneous spleen ruptures are rare. They are most commonly occurred due to infectious causes (viral, bacterial and parasitic) and hematological diseases (leukemia, lymphoma, dysglobulinemia). It is a diagnostic and therapeutic emergency. A Splenectomy hemostasis is the most common treatment [1].

Case presentation

We report the case of a 27-year-old female patient, without pathological history, admitted to the emergency service for...
the sudden onset of pain in the left hypochondrium, associated with vomiting. She had reported an influenza-like illness a week earlier for which her COVID-19 PCR was negative. On admission, she was conscious, breathing normal, pale, apyretic, mucocutaneous sub-icterus with left hypochondrium defense and moderate splenomegaly.

The ultrasound examination performed in the emergency radiology department showed a 17 cm spleen with a large heterogeneous hypoechoic subcapsular formation in the spleen, suggesting a hematoma or abscess. Peritoneal effusion was found.

Abdominal CT angiography was performed. The formation described on ultrasound corresponds to a spontaneously hyperdense subcapsular fluid collection (blood density) and not enhanced after injection of contrast agent in relation to a splenic hematoma. There are also subcapsular triangular patches, hypodense, with a peripheral base and hilar apex, which are not enhanced after injection of contrast, corresponding to foci of infarction. In addition, there is a large peritoneal effusion related to hemoperitoneum. No detectable vascular abnormalities, especially no thrombosis of splenic vein (Fig. 1).

On the basis of these clinico-radiological arguments, diagnosis of spontaneous splenic rupture was evoked.

The evolution was marked by the appearance of hemodynamic instability with signs of circulatory collapse, blood pressure 75/40 mm Hg. Hemoglobin level decreased from 12 g/dl to 8 g/dl.

Biological examinations showed: microcytic hypochromic anemia with hemoglobin at 8 g/dl, hyperleukocytosis at 16000/mm$^3$ and thrombocytopenia at 35000/mm$^3$.

The hepatic assessment indicated viral hepatitis, showing a biological cholestasis syndrome (elevated bilirubin: total bilirubin 74 mg/l, direct 62 mg/l and indirect 11 mg/l) and elevated transaminases (ALT and AST 4x normal, ALP 110 IU/L and LDH 566 IU/L).

The patient was admitted immediately to the operating room. During laparotomy, there was a large hemoperitoneum, enlarged spleen measuring 18 cm in length containing large hematoma related to complete decapsulation of spleen (Fig. 2). Splenectomy was performed with a good clinical response. The patient was released from the intensive care unit after 6 days.

An etiological work-up was initiated to look for primary tumor pathologies, hemopathies, vasculitis, coagulation disorders and above all an infectious cause (EBV, CMV, HIV, and COVID-19) or drug intake. Anti-CMV IgM elevation was identified and 6-month follow-up found positive IgG serology.

On histopathological study: The surgical specimen (splenectomy) weighs 1146 g, $18 \times 14 \times 5$ cm in size. Some areas are shredded, there is extensive capsular effraction over 15 cm and hemorrhagic parenchyma. The splenic architecture is respected with a regular white pulp, the red pulp is congestive, with hemorrhagic suffusion and necrosis. Liver biopsy showed an intra-lobular lymphocytic inflammatory infiltrate, the hepatocytes are ballooned containing bile pigments.

Medical treatment with ganciclovir was administered, with a good clinical evolution of the patient. Liver function tests was normalized. Control serology showed IgM negativation and IgG positive at 183 AU/ml (sign of previous contact with the virus).

**Discussion**

Splenic rupture is mainly caused by trauma. But in some rare cases, it can also occur without obvious trauma, known as atraumatic splenic rupture or spontaneous spleen rupture.
It is a life-threatening diagnostic and therapeutic emergency [2]. The clinical presentation can vary from left hypochondrial pain to hemorrhagic shock [3].

Three mechanisms were involved in the process: the increase in intrasplenic tension linked to cell hyperplasia and engorgement; compression by the abdominal muscles during sneezing, coughing or defecating; vascular occlusion by hyperplasia of the endothelial reticulum responsible for infarction associated or not with a subcapsular hematoma [4].

Non-traumatic ruptures of the spleen can occur in 1 or 2 phases.

Single stage rupture manifests as an acute pattern rapidly progressing to hemorrhagic shock.

In the case of a 2-stage rupture, a subcapsular hematoma is first formed. The symptomatology is sub-acute combining episodes of epigastric or left hypochondrium pain radiating to the left shoulder like in our observation. Followed by rupture of the spleen capsule [2,3,5].

A majority of non-traumatic splenic rupture has an aetiological factor leading to the rupture. They are dominated by infectious diseases (30%) represented essentially by infectious mononucleosis, malaria and bacterial endocarditis, and hematological diseases (27%) mainly hematological malignancies. More rarely, tumor pathologies, digestive pathologies such as pancreatitis or portal hypertension and renal failure at dialysis stage.

Acute leukemia and non-Hodgkin’s lymphoma are the most common hematological causes of spontaneous splenic rupture. The most likely mechanisms are: infiltration of the spleen by malignant cells; capsular distension; coagulation disorders with infarction and subcapsular hematoma [1,2,4].

Patients with spontaneous splenic rupture usually present with signs of hypovolemic shock, abdominal pain, and tenderness in the left upper quadrant. Nausea and vomiting, syncope, and vertigo are also reported among clinical symptoms. Pain may extend in the shoulder in the lying position (Kehr’s sign) secondary to blood irritation in the left hemidiaphragm. It is remarkable that Kehr’s sign has been reported to be positive in 1 out of 2 cases of spontaneous splenic rupture. However, symptoms may be atypical and the condition may imitate acute coronary ischemia, pulmonary embolism, peptic ulceration, and pneumonia [6].

Ultrasound and computed tomography have been reported to be the most useful imaging diagnostic tools. Ultrasonography can reveal an enlarged, displaced, double-countered spleen, as well as intraperitoneal bleeding. Abdominal tomography can help achieve a definitive diagnosis and enable the detection of hypodense/hyperdense foci, together with intracapsular, perirenal, and intraperitoneal fluid. Additionally, computed tomography can be used to grade the rupture [6,7].

Regardless of etiology, the immediate decisions about treatment of atraumatic splenic rupture can be varied, depending on the degree of splenic injury. If the degree of splenic injury is mild, then conservative therapy consisting of fluids, with or without blood transfusion and intensive care unit admission for close monitoring may be sufficient. If severe, then splenic artery embolization, splenic salvage, or splenectomy may be indicated when conservative management fails to achieve hemodynamic stabilization. Approximately 20% to 40% of patients require surgical intervention [7,8].

Our case presents a particular aspect. A subacute phase with pain in the left hypochondrium followed by the acute onset of hypovolemic shock in a young woman without comorbidities. On the other hand, spontaneous rupture of the spleen remains an exceptional complication of primary CMV infection.

The prevalence of CMV infection is higher the lower the socio-economic level and the higher the population density. The reservoir of the virus is strictly human. There are many asymptomatic carriers. The mechanisms of rupture are poorly understood. The role of massive lymphocytic cellular infiltration of the cell cords and vascular walls during mononucleosis syndromes, although more frequent during Epstein Barr virus infections than CMV, leading to acute ischemic phenomena at the origin of fragility of the splenic parenchyma, loss of elasticity of the capsule, and transient hemostasis disorders frequent during infectious diseases, are debated [9,10,11].
Conclusion

Spontaneous rupture of the spleen is a rare entity. It was first described in the 19th century. The mechanisms are not fully understood.

The clinical presentation is not specific and may be confused with other surgical emergencies leading to a peroperative diagnosis of splenic rupture. Spontaneous spleen rupture is a rare clinical condition that should be considered in patients hospitalized in internal medicine clinics for infectious, hematogenic, and metabolic causes, and in those with sudden abdominal pain and hypovolemia. Ultrasound and CT scans are used to establish the diagnosis in most cases. Mortality is essentially related to the delay in diagnosis and treatment and to the severity of the underlying pathology. Treatment often consists of splenectomy.

Patient consent

Written and informed consent for publication of the case was obtained from the patient.

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