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Case Report

Haemorrhagic Shock Following Spontaneous Rupture of a Cystic Lymphangioma of the Spleen - Case Report

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Abstract

Spontaneous rupture of the spleen is a rare, but potentially fatal phenomenon. The diagnosis is sometimes difficult, and often reveals an underlying pathology; however, the prognosis is highly related to early diagnosis and treatment. Cystic Lymphangioma (CL) is a benign tumor developed in the lymphatic vessels, although the splenic localization is exceptional. The isolated splenic lymphangiomas are asymptomatic and they are often incidentally discovered when symptomatic, clinical manifestations are variable and non-specific, ranging from a simple abdominal mass to complications related to the size of the spleen. Therefore, Spontaneous rupture of the spleen is an unusual complication.

We report the case of a patient admitted for intense abdominal pain with signs of acute circulatory failure, whereas abdominal ultrasound showed normal size spleen and abdominal effusion, the CT angiography guided the diagnosis by objectifying splenic infarction associated with intra and retro-peritoneal effusions. Splenectomy was urgently performed. Histopathological examination of the surgical specimen showed a congestive spleen, with cystic dilated lymphangiomatous structures of varying sizes.

Keywords: Cystic lymphangioma; circulatory failure; splenectomy

Introduction

The splenic localization of a cystic lymphangioma is an exceptional phenomenon, they are often asymptomatic and accidentally discovered, when they are symptomatic; the clinical presentation is variable and non-specific. Spontaneous rupture of a splenic lymphangioma is an unusual and potentially fatal complication. To our knowledge; only one case is described in the literature by E. Marc et al [1]. In this observation, we report the case of a patient admitted for intense abdominal pain with signs of acute circulatory failure, secondary to a spontaneous rupture of a cystic lymphangioma of the spleen.

Case Report

A 46-year-old woman, her past medical history was unremarkable, presenting to the emergency department with severe abdominal pain of sudden installation during a defecation effort. The physical examination found a patient pale, confused, collapsed; blood pressure was at 60/30 mmHg, weak peripheral pulses and cold extremities with rapid shallow breathing (35 breaths per minute). The abdomen was distended and tender, especially at the left upper quadrant. Laboratory studies showed; hemoglobin 6.8 g/dl, C-reactive protein at 12 mg / l, lactate 3.4 mmol/l, kidney function tests along with serum electrolytes were within normal limits. Given the persistence of hemodynamic instability and neurological disorders despite infusion of 1000 ml of crystalloid solution; the patient was intubated and mechanically ventilated with placement of arterial line and a triple lumen internal jugular central venous catheter, and norepinephrine started at a dose of 2 µg/kg/min. The abdominal ultrasound showed a normal-sized spleen with abundant peri-hepatic and peri-splenic effusion, and abdominal CT angiography, performed after hemodynamic optimization, revealed hemoperitoneum with intra and retro-peritoneal hematomas associated with splenic infarction (Figure 1). A laparotomy was then carried out in an emergency and found a high-abundance haemoperitoneum with a bleeding originating from the upper-internal part of a normal-sized spleen. .Haemostasis splenectomy was performed (Figure 2).

The evolution was favorable, Hemodynamic stability was restored after splenectomy and perioperative transfusion of 08 Red Blood Cells (RBC), 08 Fresh Frozen Plasma (FFP) and 6 unit of Platelet Concentrates (CPs), gradually weaned of norepinephrine and successfully extubated the next day in the intensive care unit . Histopathological examination of the resected specimen showed a congestive spleen, with cystic dilated lymphangiomatous structures of different sizes. The diagnosis was spontaneous rupture of cystic lymphangioma of the spleen.

Discussion

Spontaneous rupture of the spleen is a rare, but a serious complication of splenic tropism diseases, it was first reported by Rokitanskyin 1861 [2], and can occur in 0.1% to 0.5% of patients without Associated Trauma [3], and the age ranges from 2 to 81 years (median age = 42 years). There are signs of hemodynamic instability and shock in about 30% of the cases, during the first examination, and approximately 8% of patients die before surgery; the diagnosis is made only by an autopsy [4]. The real mechanism of the non-traumatic rupture of the spleen is not well understood yet. Three mechanisms were involved in the process: increased intra splenic tension caused by cellular hyperplasia and engorgement, compression

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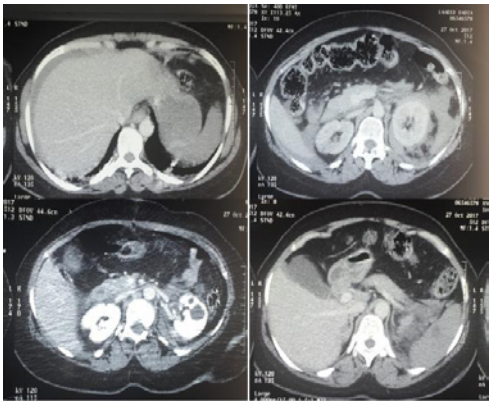

Figure 1: Axial sections of abdominal CT scan demonstrating a splenic infarction associated with intra and retroperitoneal effusions.

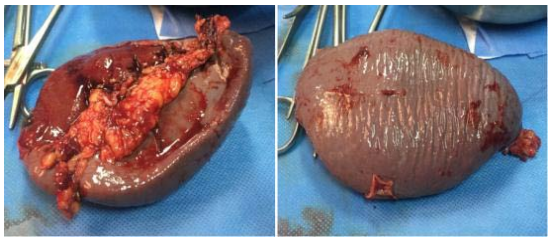

Figure 2: Normal size and appearance spleen.

by the abdominal musculature during physiological activities such as sneezing, coughing or defecation (which is the case of our patient); vascular occlusion caused by reticular endothelial hyperplasia which results in thrombosis and [5]. Swiss authors analyzed 632 publications (845 patients) concerning adults with spontaneous rupture of the spleen; the most three common etiologies involved were haematological diseases (non-Hodgkin's lymphoma), viral diseases (infectious mononucleosis) and neighborhood inflammation (pancreatitis). Which together accounted for 42%of the cases of spontaneous rupture of the spleen, and no etiology was found in 59 subjects (7%) [6].

Cystic Lymphangioma (CL) is lymphatic vessels benign congenital malformations. The splenic location of the CL is exceptional, only a few cases have been reported in the literature [7]. The neck, head and axillary regions are the common localizations. The isolated splenic lymphangiomas are usually an incidental radiological finding because patients are rarely symptomatic. When they are symptomatic, the clinical manifestations are significantly variable and non-specific; they may present as symptoms related to their size with gastric compression or pain, an abdominal mass, or infection with abscess [8]. Spontaneous rupture of the spleen is an unusual complication. In fact, in the literature, only one case recently described by E. Marc et al of spontaneous splenic rupture secondary to cystic lymphangioma in a patient with congenital factor VIII deficiency [1].

Given the lack of specificity of symptoms, the diagnosis of spontaneous splenic rupture is sometimes difficult. Where two clinical forms are conventionally observed; the subacute form characterized by left-sided abdominal pain, left shoulder pain (Kehr's sign), associated with sudden anemia, tachycardia and orthostatic hypotension. The acute form characterized by a hypovolemic shock, as was the case for our patient [9,10]. Abdominal ultrasound is the first-line examination to confirm the diagnosis. Furthermore, the abdominal CT scan has a better sensitivity for assessing the lesions [9,11]. In our case; the abdominal ultrasound showed a normal-sized spleen with presence of abdominal effusion and it is the angioscanner that allowed to guide the diagnosis by objectifying a splenic infarction associated with intra and retro-peritoneal effusions.

Surgical splenectomy is recommended as the only definitive treatment for spontaneous spleen rupture with hemodynamic instability, conservative treatment may beindicated in hemodynamically stable patients, having no clinical or scannographic arguments for a potentially progressive lesion [12,13].

Conclusion

Spontaneous splenic rupture secondary to cystic lymphangioma is an exceptional phenomenon, the current observation would be the second in the literature that illustrates the interest of early surgical exploration, even in the absence of pathognomonic signs of splenic rupture.

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