Atraumatic splenic rupture was attributed to intra-cystic haemorrhage and hypersplenism in a patient with cirrhosis and portal hypertension: A case report

SAGE Open Medical Case Reports
Volume 8: I-4
© The Author(s) 2020
Article reuse guideline:
sagepub.com/journals-permissions
DOI: 10.1177/2050313X20901900
journals.sagepub.com/home/sco



Hao Li¹, Dongyao Guan¹, Junqiang Xu¹, Enhao Jin² and Shu Sun³

Abstract

Liver cirrhosis with splenomegaly and portal hypertension has been described in the medical literature as increasing the risk of splenic rupture. We report a case of atraumatic splenic rupture in a male with liver cirrhosis associated with splenomegaly, which was further complicated by hypersplenism and intra-cystic haemorrhage in the spleen. The 56-year-old man was hospitalized because of sudden onset of intermittent pain in the left quadrant abdomen with no history of trauma. Upon admission, the patient presented no fever, palpable abdominal tenderness, splenomegaly, and hypersplenism. Enhanced computed tomography revealed a splenic subcapsular haematoma connected to a cystic mass located at the splenic hilum and free fluid in the abdomen, which is indicative of splenic rupture. The patient underwent immediate laparotomy and splenectomy followed by proper management. Post-surgery diagnostic microscopy indicated liver steatosis, perivenular fibrosis, and regenerative nodules, which are suggestive of cirrhosis. The patient was discharged from the hospital with an uneventful recovery.

Keywords

Atraumatic splenic rupture, cirrhosis, splenomegaly, hypersplenism, intra-cystic haemorrhage

Date received: 24 February 2019; accepted: 31 December 2019

Introduction

Atraumatic splenic rupture has been reported in the medical literature as having a high index of suspicion for diagnosis and requires immediate treatments to achieve a successful patient outcome. However, its mechanism, incidence, and prognosis are poorly understood due to the great heterogeneity and a limited source of reviews. It is considered to be subsequent to pathological changes in the spleen. Liver cirrhosis with splenomegaly and portal hypertension has generally been described as one of the most important pathophysiologic factors that increase the risk of splenic rupture. The substitute of the splenic rupture.

We report a case of atraumatic splenic rupture in a patient with liver cirrhosis associated with splenomegaly and portal hypertension, which was further complicated by intra-cystic haemorrhage and hypersplenism. Anatomic abnormalities in the spleen, such as large splenic cysts, hamartoma, and infarction, are also reportedly associated with splenic rupture. 8–10 However, intra-cystic haemorrhage lesions are rare in a spleen that appears to be normal or has been made

vulnerable by a preceding disease. 11,12 As such, we believe our case is special in presenting a small-sized haemorrhagic cyst in the spleen that resulted in atraumatic splenic rupture in a pre-existing condition of cirrhosis associated with splenomegaly, portal hypertension, and hypersplenism.

Case presentation

A 56-year-old male was referred to our emergency room with sudden onset of intermittent abdominal pain in the left

Department of Hepatopancreatobiliary Surgery, Affiliated Hospital of Yanbian University, Yanji, P.R. China

²Department of Imaging, Affiliated Hospital of Yanbian University, Yanji, P.R. China

³Department of Pathology, Affiliated Hospital of Yanbian University, Yanji, P.R. China

Corresponding Author:

Hao Li, Department of Hepatopancreatobiliary Surgery, Affiliated Hospital of Yanbian University, 1327 Juzi Street, Yanji 133000, Jilin, P.R. China. Email: lih@ybu.edu.cn

Creative Commons Non Commercial CC BY-NC: This article is distributed under the terms of the Creative Commons Attribution-NonCommercial 4.0 License (https://creativecommons.org/licenses/by-nc/4.0/) which permits non-commercial use, reproduction and distribution of the work without further permission provided the original work is attributed as specified on the SAGE and Open Access pages (https://us.sagepub.com/en-us/nam/open-access-at-sage).

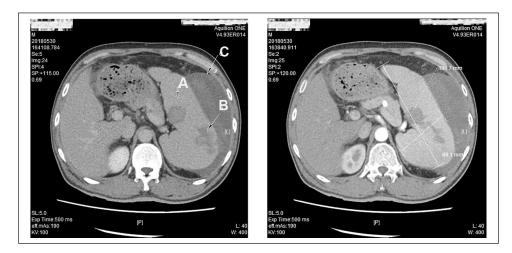


Figure 1. Abdominal computed tomography views of the patient. (Left) Arrow A: a cystic mass located at the splenic hilum and intracystic haemorrhage; Arrow B: splenic laceration; Arrow C: splenic haematoma. (Right) Intra-cystic haemorrhage leading to subcapsular haematoma.

upper quadrant. The pain started 5 days prior with no obvious traumatic cause. The patient had no clinical history of hepatitis, hypertension, coronary heart disease, diabetes, or any type of surgery, but he had a long-term history of excessive alcohol consumption with an average of 100 g of ethanol per day. During physical examination upon admission, he presented no fever with a body temperature of 36.2°C, a pulse rate of 80/min, and blood pressure of 170/90 mmHg. Abdominal palpation revealed tenderness in the left upper quadrant but no rigidity or spasm. The spleen was palpable at 10 cm below the left coastal arch. The result of the abdominal shifting dullness exam was negative. Diagnostic paracentesis yielded nonclotting blood.

The patient's laboratory tests showed a white blood cell (WBC) count of 2.03×10^9 /L, red blood cell (RBC) count of 3.46×10^9 /L, haemoglobin level (Hb) of 10^9 g/L, and platelet count of 71×10^9 /L, which are indicative of hypersplenism. Enhanced computed tomography revealed splenic subcapsular attenuation of echogenicity connected to a hypoechoic cystic mass (~3 cm) located at the splenic hilum, as well as free fluid in the abdomen, which are suggestive of intra-cystic haemorrhage—induced splenic rupture and haemoperitoneum (Figure 1).

An exploratory laparotomy was performed due to the suspicion of splenic rupture and haemoperitoneum. During the surgery, $\sim\!2300\,\mathrm{mL}$ of fluid mixed with blood clots was collected from the peritoneum. The liver exhibited an irregular or nodular surface, blunt edges, and medium hard texture, which are suggestive of liver cirrhosis. The enlarged spleen $(35\,\mathrm{cm}\times28\,\mathrm{cm})$ presented subcapsular haematoma (>40% of the surface area) and laceration (~4 cm in depth). The intraoperative portal venous pressure was measured as $35\,\mathrm{cm}\,\mathrm{H}_2\mathrm{O}$. The ruptured spleen was removed, and a liver biopsy (5 mm \times 5 mm) was obtained for histopathological purposes. During the splenectomy, the patient received a blood transfusion containing 800 mL of packed RBCs, $300\,\mathrm{mL}$ of plasma, and 1 unit of platelets.

The patient had an uneventful recovery and was discharged at 10 days post-surgery. Upon discharge, his laboratory test exhibited favourable outcomes (WBC 7.11×10^9 /L, RBC 4.87×10^9 /L, Hb $148\,\text{g/L}$, and platelets 424×10^9 /L). Post-surgery microscopy of the liver revealed macrovesicular steatosis, perivenular fibrosis, and fibrous bands subdividing the liver into regenerative nodules, which is indicative of cirrhosis (Figure 2(a) and (b)). The spleen presented intra-cystic haemorrhage and a cyst covered with an endothelial layer, but there was no calcification or fibrosis (Figure 2(c)). There was also congestion and dilation of the sinusoids, active germinal centres, and increased numbers of macrophages in the spleen pulp (Figure 2(d)).

Discussion

The majority of atraumatic splenic ruptures (over 90%) are pathologic, which is generally considered to be subsequent to pathological changes in the spleen. 13 A few cases of splenic rupture were previously reported in patients with hepatitisrelated liver cirrhosis and portal hypertension,3-7 including one rare case of spontaneous rupture of splenic hamartoma which was not encapsulated and composed of sinusoidal spaces and therefore was mostly affected by portal hypertension.3 In this case, it was apparent that the patient presented portal hypertension and severe splenomegaly due to alcoholic cirrhosis, and the laboratory test upon admission suggested typical hypersplenism. Post-surgery diagnostic microscopy also revealed active germinal centres and congestive changes in the splenic parenchyma. Moreover, computed tomography revealed subcapsular haematoma connected to intra-cystic haemorrhage. In general, nonparasitic cysts of the spleen are rare but usually benign, and non-traumatic haemorrhage in those cysts are even uncommon when the sizes are small. Surgical management has also been recommended for splenic

Li et al. 3

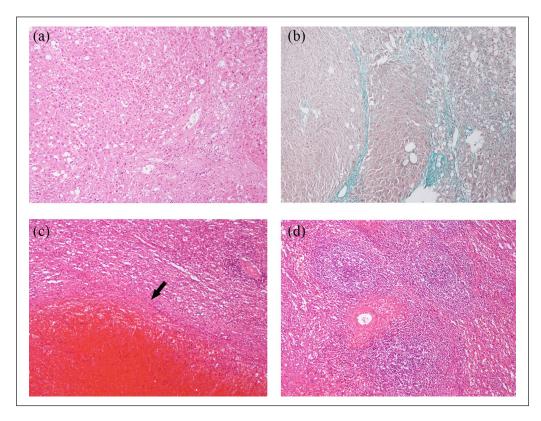


Figure 2. Pathohistological views of liver and spleen tissues from the patient. (a) The liver shows macrovesicular steatosis, H&E staining, objective, $10\times$; (b) fibrous bands subdividing liver into regenerative nodules, Trichrome staining, indicative of cirrhosis, objective, $10\times$; (c) intra-cystic haemorrhage in the spleen, H&E staining, objective, $4\times$; and (d) the spleen shows congestion and dilation of sinusoids, active germinal centres and increased numbers of macrophages, H&E staining, objective, $10\times$.

cysts with diameter larger than 4–5 cm, because they increase the risk of complications. 14,15

This case was complicated by a few unfavourable conditions that potentially increased the risk of splenic rupture. The first is the increased intra-splenic tension due to cellular hyperplasia and engorgement in splenomegaly, which may lead to splenic rupture. The spleen may be compressed by forceful diaphragmatic contraction or abdominal musculature during physiological activities, such as sneezing, coughing, and defaecation. Second, liver cirrhosis generates portal hypertension, which may give rise to splenic congestion and vascular occlusion caused by reticular endothelial hyperplasia, which in turn results in thrombosis and infarction. This is often observed in patients with splenomegaly and is involved in interstitial and subcapsular haemorrhage, as well as further stripping of the capsule. The distended capsule finally gives way and causes splenic rupture. Third, enhanced computed tomography revealed intra-cystic haemorrhage in the spleen, which may lead to subcapsular haematoma. Splenomegaly caused by severe portal hypertension is common in other patients with cirrhosis, who normally exhibit severe thrombocytopenia and leukopenia. This could worsen subcapsular haemorrhage due to coagulation dysfunction, and expanding subcapsular haematoma eventually tears the capsule. Notably, the cystic mass was located at the splenic hilum, where it increased the risk of haemorrhage due to portal venous hypertension. We suspect this to be the most direct cause of splenic rupture in our case.

Conclusion

Emergency splenectomy is required for splenic rupture. Recently, ultrasonography and computed tomography have helped improve the discovery of incidental splenic cysts. Operative indications for splenic cysts should include not only the possibility of malignant disease but also the risk of splenic rupture under certain circumstances, even when the cyst is small. We consider the pre-existing liver cirrhosis, portal hypertension, and hypersplenism to be risk factors for intra-cystic haemorrhage that eventually lead to splenic rupture.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Informed consent

Written informed consent was obtained from the patient for his anonymized medical information to be published in this article.

ORCID iD

Hao Li https://orcid.org/0000-0001-9441-6776

References

- Aubrey-Bassler FK and Sowers N. 613 cases of splenic rupture without risk factors or previously diagnosed disease: a systematic review. BMC Emerg Med 2012; 12: 11.
- Unal E, Onur MR, Akpinar E, et al. Imaging findings of splenic emergencies: a pictorial review. *Insights Imaging* 2016; 7(2): 215–222.
- Seyama Y, Tanaka N, Suzuki Y, et al. Spontaneous rupture of splenic hamartoma in a patient with hepatitis C virusrelated cirrhosis and portal hypertension: a case report and review of the literature. World J Gastroenterol 2006; 12(13): 2133–2135.
- Chien RN and Liaw YF. Pathological rupture of spleen in hepatitis B virus-related cirrhosis. Am J Gastroenterol 1993; 88(10): 1793–1795.
- Van Landingham SB, Rawls DE and Roberts JW. Pathological rupture of the spleen associated with hepatitis A. *Arch Surg* 1984; 119(2): 224–225.

- Thijs JC, Schneider AJ and van Kordelaar JM. Spontaneous rupture of the spleen complicating portal hypertension. *Intensive Care Med* 1983; 9(5): 299–300.
- 7. Wood DA. Pathologic aspects of acute epidemic hepatitis, with especial reference to early stages; report of a series of ten cases, including a case in which there was spontaneous rupture of the spleen and six cases of fulminating disease in patients who had been wounded several months previously. *Arch Pathol* 1946; 41: 345–375.
- 8. Lam CM, Yuen ST and Yuen WK. Hemoperitoneum caused by spontaneous rupture of a true splenic cyst. *Hepatogastroenterology* 1998; 45(23): 1884–1886.
- Renzulli P and Candinas D. Symptomatic retroperitoneal cyst: a diagnostic challenge. Ann R Coll Surg Engl 2009; 91(2): W9–W11.
- Al-Salem AH. Massive splenic infarction in children with sickle cell anemia and the role of splenectomy. *Pediatr Surg Int* 2013; 29(3): 281–285.
- 11. Becker MC and Brill R. Hemorrhagic cyst of the spleen in a case of Gaucher's disease. *Am J Surg* 1949; 77(1): 108–113.
- Schechter DC, Owens JC and Palmieri AJ. Hemorrhagic cyst of the spleen. Report of two cases. Am J Surg 1962; 104: 777–784.
- Tonolini M, Ierardi AM and Carrafiello G. Atraumatic splenic rupture, an underrated cause of acute abdomen. *Insights Imaging* 2016; 7: 641–646.
- Macheras A, Misiakos EP, Liakakos T, et al. Non-parasitic splenic cysts: a report of three cases. World J Gastroenterol 2005; 11(43): 6884–6887.
- Golmohammadzadeh H, Maddah G, Shams Hojjati Y, et al. Splenic cysts: analysis of 16 cases. Caspian J Intern Med 2016; 7(3): 217–221.