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Upper Gastrointestinal Bleeding Revealing Segmental Portal Hypertension Secondary to A Splenic Hydatid Cyst: An Exceptional Case

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Abstract **Case Report**

Segmental [left-sided] portal hypertension is a rare condition resulting from isolated splenic vein obstruction, most commonly secondary to pancreatic disease. Splenic hydatid cyst as an underlying cause is exceptional, with only a few cases reported. We report the case of a 75-year-old patient with a history of type 2 diabetes, hypertension, coronary artery bypass grafting on antiplatelet therapy, and prior surgery for a splenic hydatid cyst in 2016, who presented with recurrent upper gastrointestinal bleeding. The patient experienced three episodes of hematemesis associated with melena, the last occurring 15 days before admission. On examination, the patient was hemodynamically stable with marked asthenia. Upper gastrointestinal endoscopy revealed no esophageal varices, mild portal hypertensive gastropathy, and a large isolated fundal gastric varix, with red signs, successfully treated by cyanoacrylate injection; suggestive of segmental portal hypertension. Laboratory findings showed mild anemia [hemoglobin 11.1 g/dL] with preserved liver function. Abdominal ultrasonography demonstrated a 6 × 5.5 cm anechoic lesion at the splenic hilum, suggestive of a recurrent hydatid cyst or postoperative sequela causing splenic vein compression. Hydatid serology was positive. Splenic involvement accounts for only 0.5–4% of hydatid disease localizations, and its association with segmental portal hypertension remains exceedingly rare. This case highlights an unusual cause of segmental portal hypertension revealed by isolated gastric variceal bleeding and underscores the importance of considering hydatid disease in endemic regions.

Keywords: Segmental portal hypertension, Sinistral portal hypertension, Splenic hydatid cyst, Gastric varices, Splenic vein compression, Upper gastrointestinal bleeding.

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INTRODUCTION

Segmental portal hypertension, also known as left-sided portal hypertension, is an uncommon entity resulting from isolated splenic vein obstruction, leading to increased pressure in the short gastric veins and the formation of isolated gastric varices [1]. In contrast to cirrhotic portal hypertension, liver function is typically preserved and esophageal varices are absent.

The most common causes include pancreatic disorders such as chronic pancreatitis and pancreatic tumors [1]. Hydatid disease, caused by Echinococcus granulosus, remains endemic in many regions including North Africa [2]. However, splenic involvement is rare, accounting for only 0.5–4% of cases [3,4], and its complication by segmental portal hypertension is exceptional [5,6].

We report a rare case of segmental portal hypertension secondary to a splenic hydatid cyst revealed by upper gastrointestinal bleeding.

PATIENT AND OBSERVATION

A 75-year-old patient with a history of type 2 diabetes mellitus, hypertension, coronary artery bypass grafting under antiplatelet therapy, and previous surgery for a splenic hydatid cyst in 2016, presented with recurrent upper gastrointestinal bleeding.

The patient reported three episodes of hematemesis of moderate to large volume associated with melena, the most recent occurring 15 days prior to admission. No hematochezia or other digestive symptoms were noted. On examination, the patient was

hemodynamically stable, afebrile, and presented with marked asthenia.

Laboratory investigations revealed mild anemia [hemoglobin 11.1 g/dL], with normal liver function tests and coagulation parameters, and no biological signs of portal hypertension.

Esophagogastroduodenoscopy showed no esophageal varices, mild portal hypertensive gastropathy, and a large fundal gastric varix with red signs. Endoscopic treatment with cyanoacrylate injection was successfully performed without complications.

Abdominal ultrasonography demonstrated a 6 × 5.5 cm anechoic, nonvascularized lesion at the splenic hilum, compatible with a recurrent hydatid cyst or postoperative residual cavity, likely causing extrinsic compression of the splenic vein. Hydatid serology was positive.

The case was discussed in a multidisciplinary meeting. Splenectomy was initially considered; however, given the patient's advanced age and multiple comorbidities, the surgical risk was considered high. A spleen-preserving cystectomy was therefore performed without complications.

The postoperative course was favorable. Hemoglobin levels progressively normalized, and no further episodes of gastrointestinal bleeding were reported during follow-up.

DISCUSSION

Left-sided, or sinistral, portal hypertension [SPH] is an uncommon form of segmental portal hypertension caused by obstruction of splenic venous outflow. It leads to diversion of splenic blood flow through collateral pathways, particularly the short gastric and gastroepiploic veins, resulting in isolated gastric varices, often without esophageal varices or impaired liver function [1,2]. Splenic venous obstruction may result from thrombosis, stenosis, inflammation, or extrinsic compression, with pancreatic diseases being the most frequent causes [1,2].

Splenic hydatid cyst is an uncommon localization of cystic echinococcosis, which remains endemic in North Africa and the Mediterranean region. Although the liver and lungs account for most localizations, splenic involvement represents only a small proportion of cases [3,4]. Splenic cysts are often asymptomatic but may cause complications through infection, rupture, or compression of adjacent organs and vascular structures. Compression of the splenic vein by a hilar cyst is a particularly rare mechanism of SPH.

Only a few cases have described an association between splenic hydatid disease and portal hypertension. Khan *et al.,* reported a splenic hydatid cyst presenting with non-cirrhotic portal hypertension [5], while Kantarçeken *et al.,* described a patient with a splenic hydatid cyst, splenic vein compression, isolated gastric varices, and recurrent upper gastrointestinal bleeding [6]. The main characteristics of these cases and our patient are summarized in Table 1.

Table 1: Comparison of reported cases of splenic hydatid cyst associated with segmental portal hypertension

Characteristic	**Kantarçeken *et al.*, 2010**	**Khan *et al.*, 2024**	**Our case**
Country	Turkey	India	Morocco
Age / Sex	60 years / Male	32 years / Female	75 years / Male
Previous splenic hydatid surgery	No	No	Yes, 2016
Clinical presentation	Recurrent upper GI bleeding	Left hypochondrial pain; episodes of melena	Recurrent hematemesis with melena
Endoscopy	Isolated fundal gastric varices	Gastric varices	Large fundal gastric varix with red signs; no esophageal varices
Liver disease	Absent	Absent	Absent
Hydatid serology	Positive	Not clearly reported	Positive
Splenic lesion	Splenic hydatid cyst	Splenic hydatid cyst	6 × 5.5 cm hilar cystic lesion
Splenic vein involvement	Extrinsic compression	Compression	Suspected extrinsic compression
Portal hypertension	Sinistral	Non-cirrhotic	Segmental / sinistral
Initial treatment	Surgical management	Splenectomy	Cyanoacrylate injection
Definitive treatment	Cyst resection → splenectomy	Splenectomy	Spleen-preserving cystectomy
Outcome	No further bleeding after splenectomy	Favorable	No recurrent GI bleeding; hemoglobin normalized

Our case shares the characteristic features of SPH: isolated fundal gastric varix, absence of esophageal varices, preserved liver function, and a splenic hilar lesion with suspected splenic vein compression. The

positive hydatid serology and previous surgery further supported recurrent splenic hydatid disease or a postoperative residual cavity. In this setting, the combination of isolated gastric varices and preserved liver function should prompt investigation of the splenic venous system rather than assuming generalized portal hypertension [1,2].

Imaging is essential to identify the underlying cause of SPH and to assess the splenic vein, collateral circulation, and relationship between the splenic lesion and adjacent vascular structures [1,2]. In our patient, the hilar location of the cystic lesion and its suspected relationship with the splenic vein provided a plausible mechanical explanation for the segmental portal hypertension.

Management should address both the acute bleeding and its underlying cause. Cyanoacrylate injection is an established treatment for gastric variceal bleeding and successfully controlled the hemorrhage in our patient. However, endoscopic therapy does not correct the underlying splenic venous obstruction. Splenectomy has traditionally been considered definitive treatment for symptomatic SPH because it eliminates splenic venous inflow and decompresses the collateral circulation [1,6,9]. Interventional approaches, including splenic artery embolization or splenic vein stenting in selected patients, may also be considered [9].

In our patient, advanced age, cardiovascular comorbidities, antiplatelet therapy, and previous splenic surgery increased the operative risk of splenectomy. A spleen-preserving cystectomy was therefore performed to address the presumed extrinsic cause of splenic vein compression while avoiding splenectomy. The postoperative course was favorable, with normalization of hemoglobin and no recurrent gastrointestinal bleeding during follow-up. Although this outcome supports the feasibility of a spleenpreserving approach in selected patients, the limited number of reported cases precludes any definitive recommendation regarding the optimal treatment strategy.

Overall, splenic hydatid disease should be considered a rare cause of SPH in endemic regions. Isolated gastric varices in the absence of chronic liver disease, particularly when associated with a splenic hilar lesion, should prompt careful evaluation of the splenic vein and consideration of an underlying compressive lesion.

CONCLUSION

Splenic hydatid cyst is an exceptionally rare cause of left-sided portal hypertension and isolated gastric variceal bleeding. In patients presenting with isolated gastric varices, particularly in the absence of cirrhosis, esophageal varices, or impaired liver function, careful evaluation of the splenic vein and splenic hilum is essential. In endemic regions, hydatid disease should be considered among the potential causes of extrinsic splenic venous obstruction.

Although splenectomy remains an established treatment for symptomatic SPH, spleen-preserving treatment of the underlying lesion may be considered in selected high-risk patients when the mechanism of splenic venous obstruction is potentially reversible. Our case illustrates the potential role of cystectomy as a spleen-preserving strategy in a high-risk elderly patient, with favorable clinical evolution and no recurrence of gastrointestinal bleeding during followup.

Competing interests

The authors declare no competing interests.

Authors' contributions

All authors contributed to patient management and manuscript preparation. All authors read and approved the final version of the manuscript.

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