Perforated Hemocholecyst: An Unintended Consequence of Endoscopic Variceal Ligation?

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ABSTRACT

Ectopic varices are a rare sequelae of portal hypertension and present in unique ways, which may not always prompt consideration. Furthermore, endoscopic interventions on venous collaterals in the setting of portal hypertension affect the portal system hemodynamics, which may further complicate the clinical picture. We report a man with decompensated hepatitis C cirrhosis who developed hemocholecyst complicated by perforation with hemoperitoneum soon after endoscopic variceal ligation of the esophageal varices in the setting of retrospectively discovered gallbladder varices.

INTRODUCTION

Decompensated cirrhosis is defined by complications that include hepatic encephalopathy, ascites, and varices. Although most varices are located in the esophagus or gastric cardia, ectopic varices outside these locations account for approximately 5% of all variceal bleeding events.¹ Acute variceal bleeding is associated with high mortality, making surveillance and prophylaxis essential. In certain cases, primary prophylaxis against variceal bleeding involves nonselective β-blockers or endoscopic variceal ligation (EVL). Intervention, however, is not without risk and may lead to unintended consequences. Specifically, studies examining post-EVL portal pressures have demonstrated portal pressure alterations.²

CASE REPORT

A 63-year-old man with decompensated genotype 3 hepatitis C (HCV) cirrhosis presented to the emergency department (ED) with 1 day of chest pain radiating to the epigastrium, dyspnea, intractable nausea, and nonbloody emesis 10 days after EVL with variceal banding. He experienced chest pain after the procedure, but it migrated to the epigastric region the day before presentation.

His relevant medical history was significant for cirrhotic decompensations with hepatic encephalopathy and ascites. Previous treatments for his HCV, including interferon and ribavirin, were complicated by severe anemia and mood disturbances leading to therapy withdrawal. Eventual treatment with sofosbuvir and ribavirin achieved a sustained virologic response. Recently resected and irradiated seminoma precluded him from liver transplant candidacy. Years ago, he also underwent splenectomy for beta-thalassemia intermedia due to thrombocytopenia and splenomegaly; at that time, a gradient portal pressure was measured to be relatively normal and liver biopsy did not demonstrate cirrhosis.

Once diagnosed with HCV cirrhosis, he underwent variceal screening at routine intervals; medium varices were noted on previous screening. He was β-blocker intolerant because of fatigue. Ten days before presentation, surveillance esophagogastroduodenoscopy for varices demonstrated multiple columns of large varices, some with high-risk stigmata of red wale signs (Figure 1). Two bands were performed.
On arrival to the ED, he was found to be tachycardic with a normal blood pressure. Physical examination revealed jaundice, right upper quadrant and epigastric tenderness, ascites, and peripheral edema. His admission laboratory data were significant for aspartate aminotransferase of 100 U/L, alanine aminotransferase of 57 U/L, alkaline phosphatase of 246 U/L, and total bilirubin of 8.7 mg/dL with a subsequent increase to 12.8 mg/dL with direct bilirubin of 8.1 mg/dL. His international normalized ratio was 1.5, and platelets were normal.

He quickly deteriorated, developing hypotension and encephalopathy in the setting of a 2 g/dL decrease in hemoglobin. He was transferred to intensive care, and broad-spectrum antibiotics were initiated. Ultrasound with Doppler demonstrated gallbladder perforation with intracystic fluid layering and patent hepatic vasculature (Figure 2). A thoracic computed tomography angiogram—obtained in the ED for an initial concern for pulmonary embolus given his tachycardia—did not demonstrate findings concerning for cholecystitis.

Because he was a poor surgical candidate, interventional radiology was consulted for percutaneous cholecystostomy drain placement, which yielded bright red blood, as did subsequent paracentesis. Intracystic contrast injection demonstrated extravasation from the gallbladder perforation site; no contrast flowed beyond the gallbladder neck, suggesting obstruction (Figure 3). Computed tomography angiogram showed no evidence of arterial hemorrhage, leading to presumption of a venous bleed (Figure 4). A diagnosis of perforated hemocholecyst causing hemoperitoneum was made. A retrospective review of cross-sectional imaging from 1 month earlier demonstrated gallbladder varices prompting octreotide drip initiation for ectopic variceal bleeding (Figure 5). The percutaneous cholecystostomy drain remained in place for gallbladder decompression and output monitoring. A multidisciplinary team determined that the patient was not a surgical candidate, and he was ultimately transitioned to comfort care.

**DISCUSSION**

A few previous case reports have described bleeding gallbladder varices, but this case was uniquely preceded by...
a standard-of-care prophylactic procedure.\textsuperscript{3–7} Ectopic variceal bleeds are uncommon, accounting for 2%–5% of variceal bleeding.\textsuperscript{8} This case features an even rarer subset that localized to the gallbladder as ectopic varices typically lie along the primary alimentary system and rarely involve the urinary, reproductive, biliary, or gallbladder systems. The incidence of isolated gallbladder varices is estimated to be 12%–30%, most remain asymptomatic.\textsuperscript{3} There is no standard for diagnosis, and management is individualized based on best clinical judgment.\textsuperscript{9} Management options vary and include endoscopic approaches such as band ligation, transjugular intrahepatic portosystemic shunt, and balloon occluded retrograde transvenous obliteration.\textsuperscript{9}

Definitive evidence of portal hemodynamic changes post-EVL was not obtained in this patient. However, given the temporal, clinical, and radiographic correlations between bleeding and EVL, the preceding intervention was likely related.

Few studies have examined portal venous pressure changes after EVL. One study showed that in patients with a history of esophageal variceal bleeding without ascites, more than two-thirds had elevated portal pressure after EVL.\textsuperscript{2} Another study examining postprocedural imaging after EVL with sclerotherapy demonstrated that 20% of patients studied had new postprocedure nonvariceal portosystemic shunts, both suggesting portal hemodynamic changes after EVL.\textsuperscript{10}

Although the interval, in this case, was likely too short for a new shunt development, EVL may have led to increased pressure in the pre-existing varices. In the setting of pre-existing gallbladder varices and the time course of decompensation, we posit his band ligation led to altered portal hemodynamics that triggered variceal bleeding. Although no changes in portal inflow and hepatic resistance are made with EVL, the reduced venous reservoir after banding may lead to additional pressure on other varices. We suspect the increased gallbladder variceal pressure precipitated an intracystic venous bleed, particularly because

![Figure 4](image1.png)

**Figure 4.** Abdominal and thoracic computed tomography angiogram obtained after cholecystostomy showing a large hematoma inferior to right hepatic lobe (arrow) with no evidence of active arterial hemorrhage.

![Figure 5](image2.png)

**Figure 5.** Abdominal and thoracic computed tomography with intravenous and enteric contrast obtained approximately 1 month before admission. (A) Axial and (B) coronal views showing nodular enhancement within the gallbladder lumen and surrounding the gallbladder wall (arrows), compatible with intracholecystic and pericholecystic varices.
his previous splenectomy would limit collateral venous drainage. Subsequent intraluminal clot formation led to cystic duct obstruction causing hemocholecyst, and ongoing venous bleeding drove gallbladder distention and perforation, resulting in hemoperitoneum.

In the literature, hemoperitoneum secondary to gallbladder variceal bleeding in cirrhotic patients was associated with a 100% mortality rate. Timely identification and management are crucial to avoid highly morbid or fatal outcomes. Limitations partly lie in the diagnostic methods to identify hemocholecyst, as in our case where “sludge” was noted within the gallbladder that was likely blood and clot. The diagnosis of hemocholecyst was not ascertained until invasive intervention, a pattern described in the literature previously.

There are no defined management strategies for the treatment of gallbladder varices, with no clear data to direct prophylaxis. Further research is needed to answer this question more definitively.

This case describes the unusual finding of a ruptured hemocholecyst resulting from intracholecystic bleeding of ectopic gallbladder varices, observed on retrospective examination of imaging obtained before EVL. Although data are sparse, some evidence suggests that alterations to the portal system by procedures such as EVL may precipitate hemodynamic changes with secondary consequences such as the risk of bleed at sites of ectopic varices. Clinicians should be aware of the potential changes in the portal venous pressures after EVL and the subsequent potential for ectopic variceal bleeding: a rare, unintended consequence of the standard practice.

DISCLOSURES

Author contributions: MC Gasser and BW Sadowski wrote the manuscript and revised the manuscript for intellectual content. DE Baird provided the images. RM Kwok revised the manuscript for intellectual content. BW Sadowski is the article guarantor.

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