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CASE REPORT

Gastrointestinal bleeding of obscured origin due to cystic artery pseudoaneurysm



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Summary Cystic artery pseudoaneurysm is a rare condition, which usually arises from the complication of gallstone disease. Patients may present with Quinke's triad (epigastric pain, obstructive jaundice, and gastrointestinal bleeding). The results can be fatal if present with a ruptured pseudoaneurysm. We report a patient who presented with upper gastrointestinal bleeding, and later diagnosis was confirmed with a computer tomography scan of the abdomen and a three-vessel angiogram. Endovascular intervention was attempted. Although it failed, the patient was eventually cured with an open cholecystectomy.

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1. Introduction

Common presentations of cystic artery pseudoaneurysm vary from epigastric pain, obstructive jaundice, gastrointestinal bleeding, and free intraperitoneal rupture. Classically, Quinke's triad was only found in around 56% of patients.¹ Its rarity resulted in diagnostic difficulty, and mainly case reports were reported.

Management of gastrointestinal bleeding of obscured origin requires a combination of upper endoscopy and colonoscopy. However, this fails to reveal the source of bleeding in approximately 5% of patients with gastrointestinal hemorrhage.² The use of further diagnostic investigation tools depends on the stability of the patient, which include a computer tomographic (CT) scan of the abdomen, a three-vessel angiogram, red blood cell nuclear scan, small bowel enteroscopy, and operative intervention.

We present a patient who initially presented with biliary colic, and subsequently developed a complication of cystic artery pseudoaneurysm. This illustrates the management approach and timely intervention used to prevent potential complications.

Conflicts of interest: The authors disclose no conflicts of interest.

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2. Case report

A 64-year-old man presented with epigastric pain for 6 months. He was a nonsmoker and had no complaints of passing tarry stools. On physical examination, the patient was not pale and he had mild jaundice. Liver function tests showed that the serum bilirubin level was 70 $\mu\text{mol/L}$ and the alkaline phosphatase level was 224 U/L. Ultrasound of the abdomen was performed, which showed gallstones. The common bile duct (CBD) was mildly dilated with a small stone at the lower end of the CBD. Endoscopic retrograde cholangiopancreatography (ERCP) was performed on the patient which showed sludge only at the lower end of CBD. The contrast filled up the gallbladder via the cystic duct. Filling defects were found inside the gallbladder. Papillotomy was performed and an Fr 7 pigtail stent was deployed (Fig. 1). His liver function returned to normal and the patient was scheduled to receive an elective laparoscopic cholecystectomy. However, the patient complained of passing melena 10 days after the ERCP. Upper endoscopy was performed which did not reveal any bleeding from the stomach and the duodenum. The papillotomy site was clean. A colonoscopy was performed which was also normal. An urgent CT scan of the abdomen was performed (Figs. 2 and 3) which showed a lesion in the gallbladder. The noncontrast CT of the abdomen (Fig. 2) showed that there was a hypodense lesion at the gallbladder neck. There was a rim of hyperdense fluid inside the gallbladder mucosa compatible with acute hemorrhage of the gallbladder mucosa. The contrast-enhanced phase of the CT abdomen (Fig. 3) showed contrast enhancement of the gallbladder neck lesion. The overall clinical picture was compatible with cystic artery pseudoaneurysm due to complication of gallstone disease, i.e., gallstone erosion. The differential diagnosis included a benign or malignant lesion of the gallbladder leading to acute bleeding.



Figure 1 ERCP showing a normal size CBD with filling defects inside gallbladder, compatible with stones.

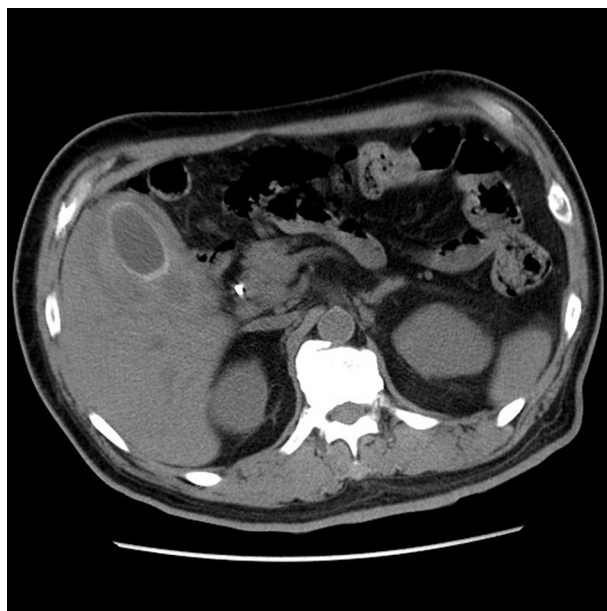


Figure 2 A noncontrast CT of the abdomen showing a hypodense lesion at the gallbladder neck.

In order to confirm the diagnosis, an urgent angiogram was performed. An angiogram including the cannulation of the celiac artery, superior mesenteric artery (SMA) and inferior mesenteric artery (IMA) was performed. Because cystic artery pseudoaneurysm was suspected, a celiac artery angiogram was performed first. Fig. 4 (Movie A) shows a celiac artery angiogram with opacification of the hepatic artery. There was no contrast extravasation in the hepatic artery and cystic artery territory. Fig. 5 (Movie B) shows an SMA angiogram. In this angiogram, there was a replaced



Figure 3 The contrast-enhanced phase of the CT scan of the abdomen, showing contrast enhancement of the gallbladder neck lesion.



Figure 4 A celiac artery angiogram with opacification of the hepatic artery. There was no contrast extravasation in the hepatic artery and cystic artery area.

right hepatic artery branching off from the SMA. Contrast extravasation could be spotted from the cystic artery supplying from the right hepatic artery.

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The diagnosis of cystic artery pseudoaneurysm was confirmed. Due to the small caliber of the cystic artery,

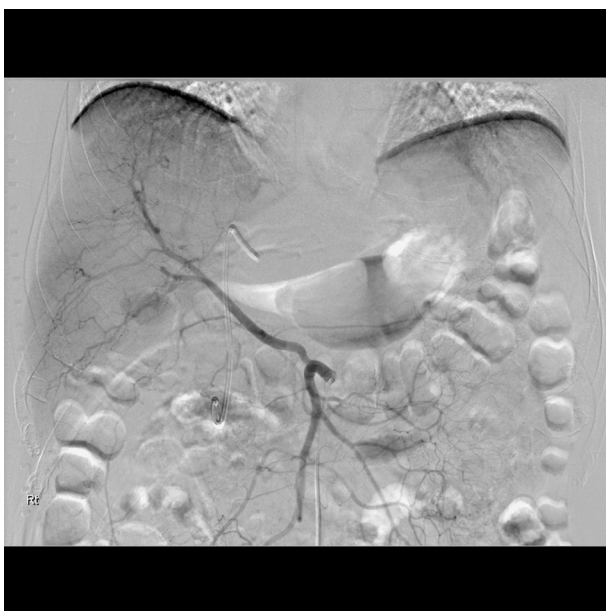


Figure 5 In this angiogram, there is a replaced right hepatic artery branching off from the SMA. Contrast extravasation can be spotted from the cystic artery supplying from the right hepatic artery.

selective cannulation and embolization of the cystic artery could not be achieved. The patient received an emergency operation. During the operation, the gallbladder was found to be fibrotic with chronic inflammation. A pseudoaneurysm of the cystic artery was located at the gallbladder neck (Fig. 6). A gallstone was found eroded through the gallbladder neck causing a pseudoaneurysm of the cystic artery. Cholecystectomy and ligation of the pseudoaneurysm was performed. The patient recovered well and was discharged 4 days after the operation. He had no further complaints of gastrointestinal bleeding.

3. Discussion

The cystic artery is a small branch of vessels from the right hepatic artery. Normal hepatic artery anatomy occurs in around 89% of the population.³ Other common variants of hepatic artery anatomy include completely replaced hepatic arterial system with a gastroduodenal artery (GDA) arising directly from the celiac axis and a replaced right hepatic artery originating from the superior mesenteric artery.⁴ It is essential that a three-vessel angiogram should be performed; otherwise a diagnosis of hemorrhage could be missed due to a variant anatomy.

Primary pseudoaneurysm of the cystic artery is rare.⁵ The precise mechanism of aneurysm formation is not well understood, although it is hypothesized that severe inflammation results in the erosion of the elastic and muscular components of the arterial wall. It may also be due to the early thrombosis of cystic artery secondary to inflammation.⁶ A patient typically presents with Quinke's triad (upper quadrant pain, obstructive jaundice, and gastrointestinal bleeding). It can lead to intermittent hemobilia mimicking gastrointestinal tract bleeding of unknown origin. Endoscopy cannot always detect the site of bleeding,⁷ which may be only detected by ERCP. An ultrasound scan of the hepatobiliary system is not an accurate modality to diagnose cystic artery pseudoaneurysm.¹ A CT scan of the abdomen remains the best method of investigation as it is easily available and noninvasive. The CT scan image of a cystic artery pseudoaneurysm, however, can be mistaken as gallstone or calcified material if not carefully interpreted. Nevertheless, despite being invasive and the

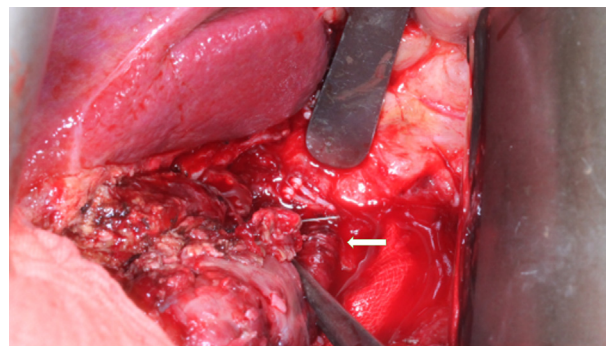


Figure 6 The gallbladder was fibrotic with chronic inflammation. A pseudoaneurysm of the cystic artery was located at the gallbladder neck.

potential risk of complications, selective hepatic angiography is still the gold standard in managing cystic artery pseudoaneurysm, for both diagnostic and therapeutic purposes.⁸

In general, cystic artery pseudoaneurysm present with hemobilia can be managed by combined endovascular and operative approaches. Transarterial embolization can temporarily stop the bleeding but may lead to local complications such as hepatobiliary necrosis, bleeding, abscess formation, and gallbladder fibrosis,⁹ catheter-related complications such as dissection, arterial–venous fistula formation, and contrast-related complications such as allergy and nephropathy. However, this offers a useful time-buying temporary measure for definitive management. In conclusion, cholecystectomy and ligation of the pseudoaneurysm provides a safe and curative way to treat this condition.

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