

Received: 2021.03.02

Accepted: 2021.05.18

Available online: 2021.06.02

Published: 2021.07.11

A Case of Massive Lower Gastrointestinal Bleed from a Cystic Artery Pseudoaneurysm Bleeding through a Cholecystocolic Fistula

Authors' Contribution:

Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Male, 66-year-old
Final Diagnosis: Cholecystocolic fistula • cystic artery pseudoaneurysm • gastrointestinal bleed
Symptoms: Hematochezia • right upper quadrant
Medication: —
Clinical Procedure: —
Specialty: Gastroenterology and Hepatology

Objective: Rare disease

Background: Acute lower gastrointestinal bleeding (GIB) is often associated with favorable outcomes. It is readily diagnosed and managed with colonoscopy, or may resolve spontaneously. Rarely, extra-colonic sources of bleeding may masquerade as lower GIB, posing a diagnostic challenge and potentially lead to harm if there are therapeutic delays. An example is cystic artery pseudoaneurysm, a rare complication of acute cholecystitis and laparoscopic cholecystectomy, which may bleed through a cholecystocolic fistula presenting as lower GIB.

Case Report: A 66-year-old man with multiple comorbidities including coronary artery disease with multiple stents and peripheral arterial disease presented with massive hematochezia. He was on aspirin 81 mg, clopidogrel 75 mg, and rivaroxaban 20 mg daily. The patient was hemodynamically unstable with BP 77/50 mmHg and heart rate 115 beats/min. Pertinent laboratory investigations showed hemoglobin 10.4 g/dL, WBC $17.2 \times 10^3/\mu\text{L}$, platelet $437 \times 10^3/\mu\text{L}$, and INR 1.28. Total bilirubin and liver enzymes were normal. Following prompt volume resuscitation with crystalloids and 2 units of O-negative blood, CT angiogram of the abdomen revealed a ruptured cystic artery pseudoaneurysm bleeding through a cholecystocolic fistula. This developed as a complication of undiagnosed gangrenous cholecystitis. The patient was sent for transcatheter embolization and the bleeding was controlled. The gangrenous cholecystitis was managed conservatively due to the patient's high surgical risk.

Conclusions: Although extra-intestinal sources of gastrointestinal bleeding are rare, clinicians should maintain a high index of suspicion, especially in elderly patients presenting concomitantly with right upper-quadrant pain. As delayed diagnosis leads to increased fatality rates, a prompt CT angiogram of the abdomen is pertinent in suspected cases.

Keywords: Gastrointestinal Hemorrhage • Intestinal Fistula • Emphysematous CholecystitisFull-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/931921>

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Background

Gastrointestinal bleeding (GIB) is a common cause of morbidity and mortality, and acute lower GIB accounts for about 20% of all cases [1]. Although most patients with lower GIB will have favorable outcomes, the prognosis is rather poor for older patients who present with hemodynamic instability and have multiple co-morbid conditions [2]. This is further worsened if the patient is actively bleeding from an unusual source, as this can result in delayed diagnosis and appropriate therapy.

In most cases of lower GIB, the source can be identified and controlled with colonoscopy as per current guidelines [1], as most bleeding originates from within the colon [3]. Rarely, the source of bleeding is outside the colon, which may pose a diagnostic challenge. To avoid potential harm, this warrants providers' awareness and a high clinical index of suspicion for prompt diagnosis and management [3-7].

Cystic artery pseudoaneurysm (CAPA) is a rare extra-colonic source of GIB. In the majority of cases, it presents as upper GIB in the form of hemobilia through the ampulla of Vater [5]. The presentation of CAPA as a lower GIB is even rarer, as this requires the formation of a fistula between the gallbladder and the large intestine.

We present a patient with massive lower GIB caused by a CAPA actively bleeding through a cholecystocolic fistula. The current literature shows that there are about 16 reported cases of CAPA with associated GIB. However, to the best of our knowledge, only 2 of these were cases of CAPA presenting as lower GIB [6,7].

Case Report

A 66-year-old man presented to the Emergency Department with heavy bright red bleeding per rectum which started suddenly, 6 h prior to presentation. He also had a 2-week history of worsening right upper-quadrant (RUQ) abdominal pain.

The patient had a past medical history of diabetes mellitus, hypertension, coronary artery disease with multiple stents, peripheral arterial disease with aorto-bifemoral bypass, and sick sinus syndrome status post pacemaker placement. His home medications included rivaroxaban 20 mg daily, aspirin 81 mg daily, and clopidogrel 75 mg daily. This was the patient's first episode of GIB. He had never undergone an upper gastrointestinal (GI) endoscopy or any biliary procedure, and a colonoscopy 3 years ago was unremarkable. There was no associated fever, nausea, or vomiting. On examination, the patient was oriented but ill-looking, pale, and anicteric. He was afebrile with a blood pressure of 77/50 mmHg, tachycardic (115

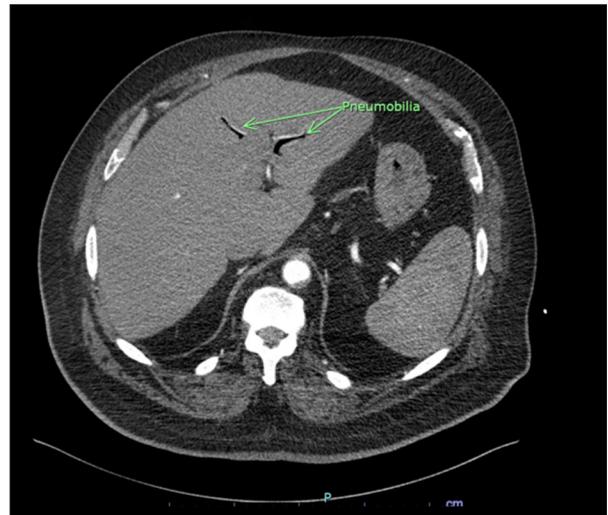


Figure 1. Axial abdominal CT angiogram showing air in the biliary tree (pneumobilia). Green arrows point to pneumobilia.

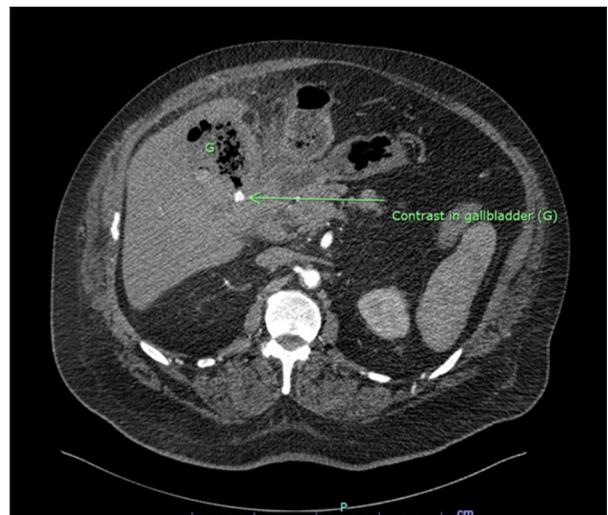


Figure 2. Axial abdominal CT angiogram showing contrast in the region of the gall bladder, suggesting ongoing bleeding.

beats/min), and tachypneic (30 cycles/min). There was diffuse abdominal tenderness, worse in the RUQ, without rebound tenderness. Murphy's sign was negative. On rectal exam, there was a significant amount of dried blood on the back and buttocks and the examining finger was stained with bright red blood.

Initial lab investigations showed a hemoglobin of 10.4 g/dL (baseline unknown), WBC $17.2 \times 10^3/\mu\text{L}$, platelet of $437 \times 10^3/\mu\text{L}$, and INR 1.28. CMP also showed creatinine of 1.46 (baseline unknown), and total bilirubin, AST, ALT, and ALP were all within normal limits. Prompt resuscitation was started with intravenous fluids and 2 units of O-negative emergency-released blood. Anticoagulants were discontinued. An abdominal computed tomography (CT) scan showed a ruptured emphysematous/



Figure 3. Pre-embolization arteriogram showing cystic artery pseudoaneurysm. A – celiac trunk; B – proper hepatic artery; C – gastrooduodenal artery; D – left hepatic artery; E – eight hepatic artery; F – cystic artery.



Figure 4. Post-embolization arteriogram with in-situ embolization coil in the region of gall bladder fossa.

gangrenous gall bladder and pneumobilia (**Figure 1**). This was concerning for a cholecystoenteric fistula. A follow-up abdominal CT angiography of the abdomen showed active arterial bleeding into the gallbladder fossa with extensive emphysematous cholecystitis and a cholecystocolic fistula with the hepatic flexure of the colon (**Figure 2**).

After initial resuscitation, the patient underwent emergency arterial transcatheter embolization, and a large pseudoaneurysm of the cystic artery was seen in the gallbladder fossa (**Figure 3**). The target cystic artery was successfully occluded with sub-selective coil embolization and the patency of the proximal non-target branch vessel was preserved (**Figure 4**).

Upper and lower GI endoscopies were performed once the patient was stable. Upper GI endoscopy showed edematous mucosa in the duodenal bulb and a similar finding was noted in hepatic flexure of colon with areas of ecchymosis in the latter. Biopsies obtained showed non-specific inflammatory changes in the duodenum and colon. Given his extensive medical comorbidities, his calculated revised cardiac risk index was 4 (class IV). This correlated to a 15% 30-day risk of death, myocardial infarction, or cardiac arrest post-surgery. As the patient was considered a high surgical risk, and the source of the GIB had been controlled, the gangrenous cholecystitis was treated conservatively with antibiotics. The patient responded well to treatment, with no further episodes of GI bleed, and he was discharged home after a 6-day hospital stay.

Follow-up abdominal CT imaging obtained 6 weeks after discharge showed a significantly contracted gall bladder, and at 16 weeks the gallbladder was not visible on imaging. He was back to his baseline health and remained clinically stable.

Discussion

CAPAs are uncommon and mostly occur as a complication of biliary tract procedures such as laparoscopic cholecystectomy or gallbladder inflammation [7,8]. The visceral inflammation extends to the arterial adventitia leading to thrombosis of the vasa vasorum, localized blood vessel wall weakness, and predisposition to rupture [5]. Cholecystocolic fistulas may arise as a complication of chronic gallbladder inflammation when pressure from the nearby enteric lumen facilitates lumen wall erosion by underlying gallstones. In our patient, the underlying gangrenous cholecystitis was the most likely predisposing factor for fistula formation. This is similar to the case described by Carey et al and Lee et al, where the patients had underlying cholecystitis [6,7]. Ostiz et al also described a patient with underlying chronic gallstone disease who presented with massive lower GIB through a cholecystocolic fistula, although the exact bleeding source was not localized [9]. As in our case, these patients were mostly older adults with multiple medical comorbidities.

CAPAs have varied clinical presentations. They are mostly asymptomatic and discovered incidentally on imaging [8,10]. However, in the event of rupture, CAPAs may present as upper GIB with hemobilia [5], sometimes associated with RUQ abdominal pain and jaundice, forming the classic Quincke triad [8,11]. While most CAPAs are associated with biliary tract instrumentation, CAPAs occurring concomitantly with cholecystoenteric fistulas are associated with underlying gall bladder inflammation [6,7,9,12-14], suggesting a central role of cholecystitis in the pathophysiology. The diagnosis of CAPA is based on imaging [15]. Although conventional angiography is the criterion

standard, its use is limited by its invasiveness. Abdominal CT angiography is the preferred initial imaging modality, as it is sensitive, non-invasive, and can also identify underlying gallbladder disease [7,15]. Hepatic ultrasound is cheaper but has a low sensitivity and the image quality may be distorted by the acoustic shadowing of gall bladder calculi [15].

Currently, there are no standard guidelines for the management of CAPA. Treatment options include open surgical repair, laparoscopic repair, and selective arterial embolization [8]. Selective embolization has a 75% success rate, with complications occurring in less than 2% of patients [16]. Post-embolization complications include ischemic hepatitis and abscess formation. None of these complications were seen in our patient. It is recommended that for CAPAs that occur secondary to gallbladder pathology, a follow-up cholecystectomy is performed once the patient is clinically stable. However, as in our patient, management may be conservative with antibiotics if the risk of surgery is deemed too high due to underlying comorbidities.

References:

1. Strate LL, Gralnek IM. ACG Clinical Guideline: Management of patients with acute lower gastrointestinal bleeding. *Am J Gastroenterol*. 2016;111(4):459-74
2. Strate LL, Ayanian JZ, Kotler G, Syngal S. Risk factors for mortality in lower intestinal bleeding. *Clin Gastroenterol Hepatol*. 2008;6(9):1004-10; quiz 955
3. Charilaou P, Devani K, Enjamuri D, et al. Epidemiology of lower GI bleed in the United States – an update from the national inpatient survey 2005-2014: Abstract #559. *Am J Gastroenterol* 2018; 113(Suppl.): S319-S321
4. Liu B, Lewis AR, Ward TJ. Cystic artery pseudoaneurysm. *J Vasc Interv Radiol*. 2016;27(5):694
5. Saluja SS, Ray S, Gulati MS, et al. Acute cholecystitis with massive upper gastrointestinal bleed: a case report and review of the literature. *BMC Gastroenterol*. 2007;7:12
6. Lee JW, Kim MY, Kim YJ, Suh CH. CT of acute lower GI bleeding in chronic cholecystitis: concomitant pseudoaneurysm of cystic artery and cholecystocolonic fistula. *Clin Radiol*. 2006;61(7):634-36
7. Carey F, Rault M, Crawford M, et al. Case report: Cystic artery pseudoaneurysm presenting as a massive per rectum bleed treated with percutaneous coil embolization. *CVIR Endovasc*. 2020;3(1):8
8. Loizides S, Ali A, Newton R, Singh KK. Laparoscopic management of a cystic artery pseudoaneurysm in a patient with calculus cholecystitis. *Int J Surg Case Rep*. 2015;14:182-85
9. Ostiz M, Vila JJ, García Sancho J, et al. [Cholecystocolic fistula: An uncommon cause of lower gastrointestinal bleeding]. *An Sist Sanit Navar*. 2012;35(3):5115
10. Nkwam N, Heppenstall K. Unruptured pseudoaneurysm of the cystic artery associated with acute calculus cholecystitis. *J Surg Case Rep*. 2010;2010(2):4
11. Praveen Kumar Sunkara PRV, Shah PK, Rakshit K, et al. Rupture of cystic artery pseudoaneurysm: A rare complication of acute cholecystitis. *Indian J Surg*. 2018;80(1):87-89
12. Changku J, Shaohua S, Zhicheng Z, Shusen Z. Upper gastric tract bleeding due to cholecystoduodenal fistula: A case report. *Hepatogastroenterology*. 2005;52(65):1372-74
13. Vadioaloo DK, Loo GH, Leow VM, Subramaniam M. Massive upper gastrointestinal bleeding: A rare complication of cholecystoduodenal fistula. *BMJ Case Rep*. 2019;12(5):e228654
14. Lee SB, Ryu KH, Ryu JK, et al. Acute acalculous cholecystitis associated with cholecystoduodenal fistula and duodenal bleeding. A case report. *Korean J Intern Med*. 2003;18(2):109-14
15. Chong JJ, O'Connell T, Munk PL, et al. Case of the month #176: Pseudoaneurysm of the cystic artery. *Can Assoc Radiol J*. 2012;63(2):153-55
16. Trombatore C, Scilletta R, Bellavia N, et al. Acute hemobilia from a pseudoaneurysm of the cystic artery arising from the left hepatic artery: Case report and literature review. *Int J Surg Case Rep*. 2017;37:60-64

Conclusions

CAPA bleeding through cholecystoenteric fistula is an uncommon but potentially fatal source of GIB if not promptly diagnosed and treated. While investigating the more common causes of GIB, clinicians should maintain a high index of suspicion in patients presenting with acute GIB and RUQ pain, or in those with a history of gallbladder disease. An abdominal CT angiogram is useful if a bleeding CAPA is suspected, and a multidisciplinary approach to management is key to improve patient outcomes.

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