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Multidisciplinary Management of Upper Gastrointestinal Bleeding Secondary to Cystic Artery Pseudoaneurysm

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Abstract

Cystic Artery Pseudoaneurysms (CAPs) are rare but have been associated with a high risk of complications including hemorrhage. While most often associated with cholecystitis, the presentation is inconsistent and management remains controversial. Similarly, the diagnostic workup is variable and typically includes endoscopy, computed tomography and angiography. Traditionally, angioembolization followed by open cholecystectomy has been the most common treatment approach; however laparoscopic management and selective pseudoaneurysm embolization alone have also been reported. We present an interesting case of cholelithiasis and choledocholithiasis associated with an upper gastrointestinal bleed secondary to a CAP which was successfully managed with a staged multidisciplinary approach.

Keywords: Cystic artery pseudoaneurysm • Gallbladder • Cholecystitis • Choledocholithiasis

Introduction

Cystic Artery Pseudoaneurysms (CAPs) are a rare occurrence with a high risk of associated complications, with only 67 cases reported from 1991 to 2020 [1]. CAPs are most associated with cholecystitis, with other contributory factors including cholelithiasis, pancreatitis and following procedures such as cholecystectomy [2]. In the absence of rupture, CAPs are typically discovered incidentally on imaging. Rupture can lead to hemoperitoneum and erosion into the biliary system is associated with biliary obstruction, haemobilia and gastrointestinal bleeding. Diagnostic modalities reported in the CAP workup include endoscopy, Computed Tomography (CT) and angiography [3]. To date, the management of CAPs remains variable. Treatment typically involves a combination of endovascular embolization, open versus laparoscopic cystic artery ligation and cholecystectomy [1,4].

We present a case of cholelithiasis and choledocholithiasis with a CAP causing an acute upper Gastrointestinal (GI) bleed. This patient was successfully managed at a tertiary care facility with a staged multidisciplinary approach involving Interventional Radiology (IR), an experienced surgical team and Gastroenterology with advanced endoscopic capabilities.

Case Report

A 74-year male presented to his local Emergency Department with a recurrent episode of right upper quadrant abdominal pain. Workup demonstrated cholecystitis and an elevated bilirubin of 10 mg/dl concerning for choledocholithiasis. He was admitted and Endoscopic Retrograde Cholangiopancreatography (ERCP) was attempted twice but unsuccessful due to copious amounts of blood in the stomach. He required two units of packed red blood cell transfusion and CT angiogram was performed demonstrating a cystic artery originating off a replaced right hepatic artery with a 1 cm CAP, Figure 1. There was no active extravasation. He was started on piperacillin-tazobactam for empiric coverage against ascending cholangitis and transferred to our institution.

He then underwent angiography and coil embolization of his CAP, Figure 2. He was subsequently taken to the operating room. Through a right subcostal incision, dense adhesions from colon were taken down revealing a thickened, distended gallbladder. The dome of the gallbladder was incised to allow removal of two large gallstones. The embolization coil was retrieved from the lumen of the gallbladder, confirming a fistula from the pseudoaneurysm into the gallbladder causing his initial upper GI bleed. There was no active bleeding after removal of the coil. The portal triad was dissected to identify the replaced right hepatic artery which was dissected free from the infundibulum of the gallbladder. The CAP to gallbladder fistula and cystic artery at the origin of the right hepatic artery were ligated. There was an easily palpable pulse in the replaced right hepatic artery. The gallbladder was then dissected free from the liver requiring resection of two portions of segment 4B due to chronic inflammation obscuring the dissection plane. Once mobilized, an intraoperative cholangiogram was performed with the cholangiogram catheter through the lumen of the gallbladder. Two opacities were seen within the distal common bile duct. The duct was flushed with saline and two large pieces of mucin and clot were retrieved from the distal common bile duct. Subsequent cholangiogram demonstrated no further filling defect with the common bile duct or hepatic ducts. The gallbladder was resected at the short cystic duct stump and a drain was placed.

On postoperative day three, bile was noted in his surgical drain prompting further workup. Repeat CT scan demonstrated no intra or extrahepatic biliary ductal dilation or undrained fluid collections. The following day, he underwent ERCP which did not demonstrate any abnormal findings within the biliary tree or extravasation from the cystic duct stump and a stent was placed in the common bile duct. His bilious drain output resolved and his serum bilirubin levels down trended with resolution of his jaundice. His drain was removed and he continues to recover well in the outpatient setting. Final pathology was negative for any dysplasia or malignancy.

Discussion

We report a case of cholelithiasis and choledocholithiasis with a CAP causing an acute upper GI bleed, which was successfully managed in a staged fashion with a multidisciplinary team.

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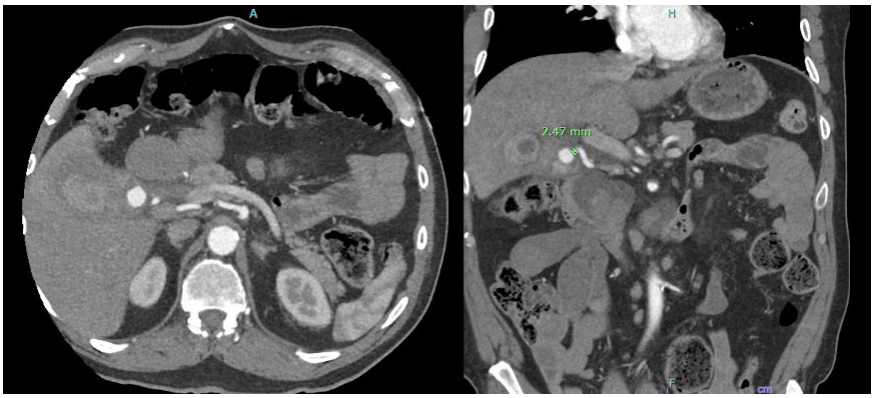

Figure 1. Preoperative CT angiogram demonstrating cystic artery pseudoaneurysm, axial view (left) and coronal view (right).

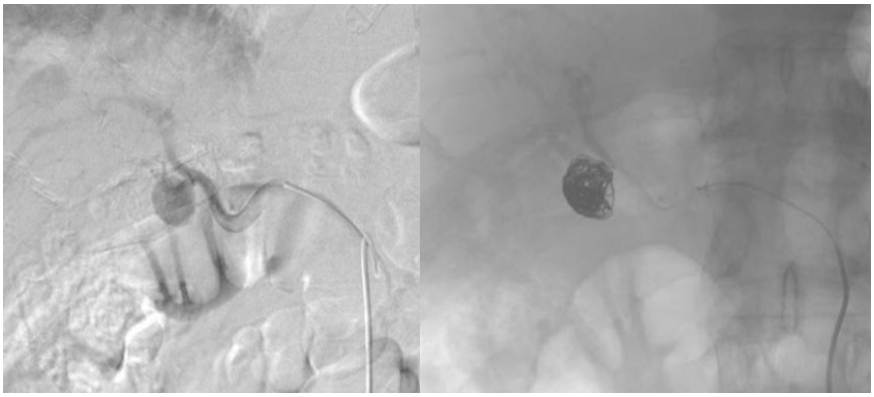

Figure 2. Angiogram of cystic artery pseudoaneurysm (left), angiogram post coil embolization (right).

CAP remain a rare occurrence; while classically associated with Quinke's triad of jaundice, right upper quadrant abdominal pain and upper GI bleeding, the presentation is often more heterogenous [1]. Common diagnostic modalities including CT and angiography were utilized in our case to confirm the diagnosis of CAP and help plan for successful management.

Given the rare nature of CAP, management strategies remain controversial [5]. Siddiqui, et al. reported a case which was similarly managed with IR embolization of the pseudoaneurysm followed by open cholecystectomy [6]. Others have reported successful management with laparoscopic approach [4]. While an open approach has traditionally been the most common due to risk of pseudoaneurysm rupture intraoperatively, these authors demonstrate laparoscopic management can be performed in select cases with favorable anatomy. In our case, the degree of inflammation and extensive dissection required around the gallbladder precluded a safe laparoscopic approach.

Meanwhile, some have also reported positive outcomes in cases of pseudoaneurysm embolization alone without cholecystectomy. This approach is often utilized in high-risk patients who are not fit to undergo surgery and commonly involves selective microembolization with close follow-up. This remains controversial however due to the increased risk of gallbladder necrosis [5].

Ultimately, diagnosis of CAP requires a high index of suspicion. We recommend liberal use of various imaging modalities as appropriate and utilization of subspecialty services for assistance with management. While some have reported on pseudoaneurysm embolization alone, we proceeded with staged cholecystectomy in the setting of cholecystitis, cholelithiasis and choledocholithiasis.

Acknowledgement

None.

Conflict of Interest

None.

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