



Cystic Artery Pseudoaneurysm with Concurrent Cholecystoduodenal Fistula—Endovascular Management and Review of Literature

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J Clin Interv Radiol ISVIR 2023;7:57–59.

Abstract

Keywords

- ▶ cystic artery pseudoaneurysm
- ▶ angioembolization
- ▶ *n*-BCA glue

A rare complication of acute cholecystitis is a pseudoaneurysm of the cystic artery. We discuss a case of a 65-year-old man with synchronous cholecystoenteric fistula and cystic artery pseudoaneurysm managed by selective angioembolization of the cystic artery, followed by interval cholecystectomy.

Introduction

Pseudoaneurysm of the cystic artery is a rare phenomenon and majority of these are secondary to inflammatory conditions of the gallbladder (GB). Synchronous presence of cholecystoenteric fistula and cystic artery pseudoaneurysm is a rare occurrence.

Case Report

A 65-year-old man was hospitalized after experiencing hematemesis, fever, jaundice, and black stools for the last 7 days. His laboratory investigations revealed low hemoglobin (7.5 g/dL), increased total leukocyte count (16,500/mm³), and conjugated hyperbilirubinemia (total bilirubin: 4.3 mg/dL). Ultrasonography (USG) showed mild bilobar intrahepatic biliary radicals dilatation. GB was distended with shaggy wall and GB calculus. Upper gastrointestinal endoscopy revealed heavy blood clots in the antropyloric region. On computed tomography (CT) scan, hyperdense contents were noted in the GB and the duodenum

(▶ Fig. 1A). The arterial phase revealed a 9 × 7 mm cystic artery pseudoaneurysm (▶ Fig. 1B, D). Air foci were noted in GB lumen with the presence of cholecystoduodenal fistula (▶ Fig. 1B, C). Although open cholecystectomy along with fistula repair would have been the treatment of choice, endovascular management was chosen in view of ongoing sepsis and bleeding. A digital subtraction angiography was performed (▶ Fig. 2). The cystic artery was superselectively catheterized on a selective angiographic run, and a 9-mm pseudoaneurysm was noted from the cystic artery (▶ Fig. 2A). Attempts were made to selectively advance the microcatheter distal to the pseudoaneurysm to embolize the distal collaterals (to avoid retrograde filling of the pseudoaneurysm), but despite repeated attempts, the microcatheter could not be placed distally (▶ Fig. 2B). Due to the presence of distal collaterals and an expendable artery, the pseudoaneurysm was embolized with a 50% *n*-butyl cyanoacrylate (*n*-BCA) glue–Lipiodol mixture (0.3 mL). Glue cast filled the pseudoaneurysm as well as the distal branches and the proximal cystic artery (▶ Fig. 2C, D). The patient had no further bleeding episodes and underwent open cholecystectomy later.

article published online
March 9, 2022

DOI <https://doi.org/10.1055/s-0042-1745919>.
ISSN 2457-0214.

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Fig. 1 A 65-year-old male with a cystic artery pseudoaneurysm. Axial noncontrast computed tomography (A) showing hyperdense content in the gallbladder (dashed black arrow) and duodenum (asterisk). Coronal arterial phase (B) showing cholecystoduodenal fistula (white arrow) and pseudoaneurysm (black arrow). Portal venous phase showing (C) thick-walled gallbladder containing air foci within and pseudoaneurysm (black arrow). Coronal maximum intensity projection (D) showing pseudoaneurysm arising from the cystic artery (black arrow).

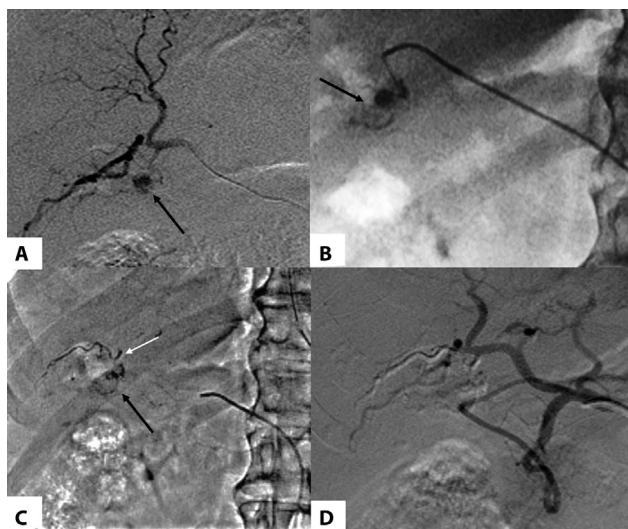


Fig. 2 A 65-year-old male with a cystic artery pseudoaneurysm. Digital subtraction angiogram performed via celiac axis injection. Selective run from right hepatic artery (A) shows pseudoaneurysm arising from the cystic artery (black arrow). Selective cannulation of the cystic artery (B) showing pseudoaneurysm (black arrow). Post-embolization (C) showing glue cast filling the pseudoaneurysm and the distal collateral (black arrow) and the proximal stump (white arrow). Postembolization angiographic run image (D) illustrating successful treatment of the cystic artery pseudoaneurysm without filling of the pseudoaneurysm.

Discussion

Cystic artery pseudoaneurysm and cholecystoenteric fistulae are two rare complications of gallstone disease (GSD).¹ Acute inflammation caused by gallstones has resulted in the

creation of a cholecystoduodenal fistula along with a cystic artery pseudoaneurysm in this case. Unique to this case was the concurrent occurrence of obstructive jaundice, cholangitis, pseudoaneurysm, and cholecystoduodenal fistula. All of these are uncommon complications of cholecystitis and their co-occurrence is even more unusual.

Cystic artery pseudoaneurysm is a rare entity. Laparoscopic cholecystectomy is the most common cause, followed by acute cholecystitis.²

The inflammatory process disrupts the artery's adventitia and promotes vasa vasorum thrombosis, which leads to media and intima erosion and, eventually, the formation of a pseudoaneurysm.³

Cholecystoenteric fistula is a rare and late complication of GSD, with a reported incidence of 0.5 to 0.9%.² Cholecystoduodenal fistula is the most common type (75–80%).³ Obstruction of the cystic duct causes repeated inflammation, causing the GB to adhere to an adjacent viscus, in this case the duodenum, causing erosion and, eventually, cholecystoenteric fistula formation. GB wall necrosis caused by mechanical pressure from an impacted gallstone can also cause fistula formation.² A ruptured cystic artery pseudoaneurysm may present as a gastrointestinal hemorrhage or hemorrhagic cholecystitis (rupture of pseudoaneurysm within the wall and into the lumen) or as intraperitoneal hemorrhage (pseudoaneurysm outside wall).^{1,4}

Our patient presented with upper abdomen pain, obstructive jaundice, and gastrointestinal bleed. This classic triad is known as Quincke's triad and is seen in ~56% of the cases with haemobilia

On USG, ruptured pseudoaneurysm may show echogenic content in the GB. "Yin-Yang" flow pattern may be noted in the pseudoaneurysm on color Doppler. Multiphasic CT is the most sensitive and widely used modality. It not only demonstrates pseudoaneurysm but also the underlying cause. CT also helps in endovascular embolization planning.⁴

There are currently no clear guidelines for the management of cystic artery pseudoaneurysm due to its rarity. Percutaneous selective cystic artery embolization is an effective treatment option in acute settings with higher rates of hemostasis. It has shown reduced mortality and morbidity than surgery in the acute setting.⁴ A two-stage approach may be used, with embolization of the pseudoaneurysm first to stabilize the patient, followed by surgery once the patient is stable.^{2,3}

Gelfoam, coils, and *n*-BCA glue all have been used for embolization.¹ Coil embolization is the most widely used technique. The origin of the pseudoaneurysm is sandwiched using coils, which cut off both the antegrade as well as retrograde flow to the pseudoaneurysm. In some cases, crossing the pseudoaneurysm and blocking the "back door access" may be difficult; in these cases, liquid embolization agents like *n*-BCA glue may be useful. This was the reason we used glue in our case. However, liquid embolic agents, such as glue, may increase the pressure within the pseudoaneurysm and increase the chance of rupture; therefore, they must be used in calculated quantities.⁵ It is worth noting that while cholecystectomy with cystic artery ligation is the definitive treatment for cystic

artery pseudoaneurysm, secondary to inflammatory conditions, Angioembolisation may have to be done as an initial lifesaving procedure for unstable patients.

Conflict of Interest

None declared.

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