

## CASE REPORT

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# Perforated gallbladder after cystic artery pseudoaneurysm embolization: A case report and review of the literature

Ramy Ahmed, Abdelrahman Omar

## ABSTRACT

**Introduction:** Cystic artery pseudoaneurysm (CAP) is an uncommon complication of cholecystitis. Transarterial embolization is an effective treatment for CAP rupture, but it carries a theoretical risk of gallbladder (GB) ischemia, which is rarely reported.

**Case Report:** A 75-year-old diabetic woman who presented with acute calculous cholecystitis complicated by CAP rupture, manifesting as massive hematemesis and melena. Urgent angiography demonstrated a large cystic artery pseudoaneurysm, which was successfully embolized using an NBCA–Lipiodol mixture. One week after embolization, the patient developed fever and right upper quadrant pain. Imaging revealed a perforated, gangrenous gallbladder with a large subhepatic abscess. As the patient was a poor surgical candidate, percutaneous transhepatic drainage was performed, resulting in complete resolution of the abscess and clinical recovery. This case highlights that the fear of gallbladder perforation should not contraindicate life-saving cystic artery embolization, as it remains amenable to minimally invasive treatment.

**Conclusion:** Gallbladder ischemia and perforation following cystic artery embolization are rare but important complications. In surgically unfit patients, percutaneous catheter drainage can provide an effective, minimally invasive alternative to cholecystectomy.

**Keywords:** Cystic artery pseudoaneurysm, Gallbladder perforation, Percutaneous drainage, Transarterial embolization

### How to cite this article

Ahmed R, Ahmed A. Perforated gallbladder after cystic artery pseudoaneurysm embolization: A case report and review of the literature. *J Case Rep Images Surg* 2026;12(1):20–24.

Article ID: 100169Z12RA2026

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doi: 10.5348/100169Z12RA2026CR

## INTRODUCTION

Cystic artery pseudoaneurysm (CAP) is a very rare complication of cholecystitis and cholecystectomy [1]. Cystic artery pseudoaneurysm rupture may present with a wide spectrum of severity, ranging from mild anemia to life-threatening hemorrhagic shock due to hematemesis, melena, or hemoperitoneum [2]. Management of cholecystitis-related CAP rupture traditionally includes urgent cholecystectomy with aneurysm resection [3] or transarterial embolization followed by elective cholecystectomy to reduce the risk of gallbladder (GB) ischemia [2].

Although GB ischemia is considered the most feared complication after cystic artery embolization, it is rarely reported in the literature, likely due to collateral perfusion from the epicholedochal arterial plexus [4–6].

We report a case of CAP rupture treated with *n*-Butyl cyanoacrylate (NBCA) embolization, complicated by GB ischemia and perforation, successfully managed with percutaneous drainage in a surgically unfit patient.

## CASE REPORT

A 75-year-old diabetic woman presented with upper abdominal pain and jaundice. Ultrasound revealed

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Received: 04 April 2026

Accepted: 19 May 2026

Published: 19 June 2026

a distended gallbladder with echogenic content, wall edema, and pericholecystic fluid. Magnetic resonance cholangiopancreatography (MRCP) (Figure 1) demonstrated a dilated common bile duct (CBD) containing signal-void material initially interpreted as stones or biliary sludge.

On admission, the patient was hemodynamically stable but anemic (hemoglobin 7 g/dL) with leukocytosis and elevated C-reactive protein (CRP). Liver enzymes and coagulation profile were within acceptable limits.

The following day, she developed massive melena with a hemoglobin drop to 5 g/dL and hypotension. After resuscitation and blood transfusion, urgent conventional angiography was performed. Celiac angiography (Figure 2) using 5F Cobra catheter shows large pseudoaneurysm arising from the cystic artery with no detected active bleeding from the aneurysm. Selective catheterization of the cystic artery was done using microcatheter (Renegade HI-FLO, Boston-scientific; Ireland). The cystic artery and the pseudoaneurysm were embolized using NBCA: Lipiodol mixture (1:1). During microcatheter withdrawal from the cystic artery after NBCA injection, fragments of the NBCA refluxed back into the segmental branches of the right hepatic artery. Post-embolization angiogram showed complete embolization of the cystic artery and pseudoaneurysm with patent both left and right hepatic arteries. The patient stabilized clinically with no further bleeding.

One week after embolization, the patient developed right hypochondrial and epigastric pain and fever. Abdominal ultrasound (US) and computed tomography (CT) (Figure 3) showed large, localized abscess in the subhepatic region related to the GB region. Perforated gangrenous GB with subhepatic abscess formation

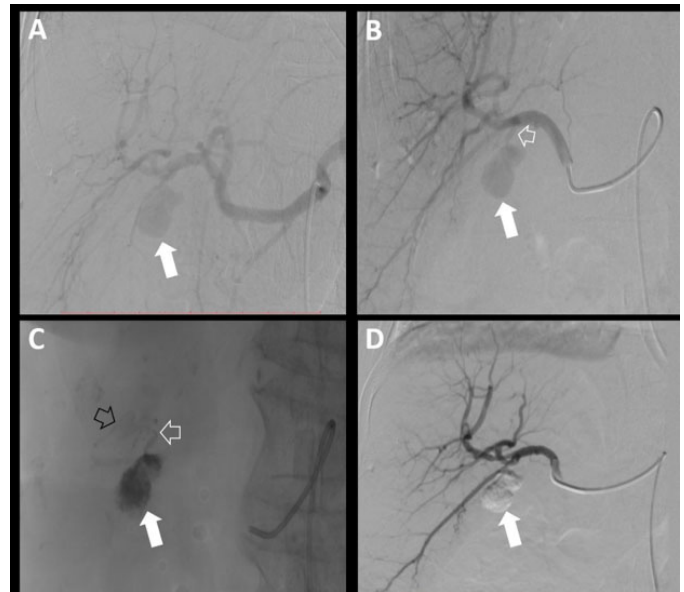


Figure 2: Conventional angiography and trans-arterial embolization. (A) and (B) Celiac and Rt hepatic arteries digital subtraction angiography (DSA) image shows large pseudoaneurysm (hollow arrow) arising from cystic artery (hollow arrow). (C) and (D) Post-embolization fluoroscopic image and Rt hepatic angiography showing complete occlusion of the cystic artery (hollow white arrow), the pseudoaneurysm (solid arrow) with NBCA: lipiodol mixture and small right hepatic artery sub-segmental branches (hollow black arrow) and patent hepatic artery.

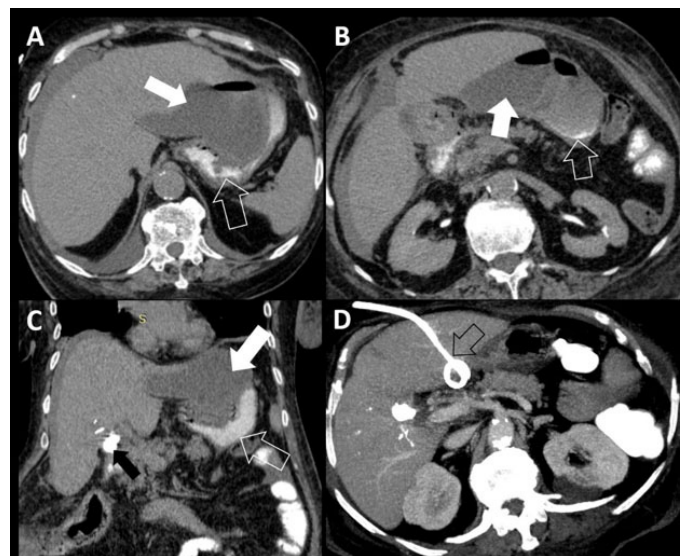


Figure 3: Post-embolization CT abdomen. (A)–(C) CT images 1-week post-embolization show large subhepatic abscess (white solid arrow) compressing upon the contrast filled stomach (hollow white arrow). Note the NBCA/Lipiodol cast within CAP (solid black arrow). (D) Follow-up CT shows complete drainage of the subhepatic abscess after percutaneous transhepatic pig tail catheter placement (hollow black arrow).

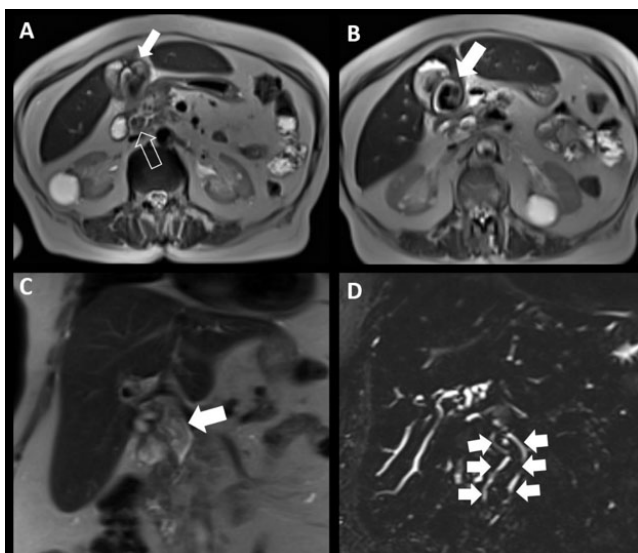


Figure 1: MRI abdomen. (A) and (B) Axial MRI showing hypointense signal lesion within the CBD (hollow arrow) and GB (solid arrow) interpreted as CBD and GB stones. (C) and (D) Coronal MRI showing intermediate signal intensity diffuse lesion filling the whole length of the CBD (solid arrows) interpreted as biliary mud. Retrospectively, these lesions are consistent with blood clots after CAP rupture.

was diagnosed. Surgical consultation recommended percutaneous drainage over surgical intervention due to patient poor general conditions. Urgent percutaneous transhepatic drainage of the abscess was done under US guidance. Follow-up CT performed 10 days after

Pig-tail catheter insertion in the abscess cavity showed complete drainage of the subhepatic abscess. Pig-tail catheter was removed, and the patient was discharged with Hgb level 10 mg/dL, white blood cells (WBCs)  $8.3 \times 10^9/L$ , alanine aminotransferase (ALT) 8 IU/L, aspartate aminotransferase (AST) 27 IU/L, alkaline phosphatase (ALP) 227 IU/L, Bilirubin 0.3 mg/dL (Total) and 0.2 mg/dL (Direct), and CRP 3 mg/L.

## DISCUSSION

Cystic artery pseudoaneurysm is a rare complication of cholecystitis, cholecystectomy and pancreatitis [1]. Up to our best knowledge, about 96 cases of CAP were reported in the English literature from 1983 to 2020. About 72% of them were associated with cholecystitis (either calcular or acalcular) where it is believed to develop from erosion of cystic artery wall by the inflammatory process and/or traumatic injury caused by GB stones [7]. Arterial wall weakness due to sclerotic changes associated with hypertension and diabetes may predispose for pseudoaneurysm formation [6]. Early thrombosis of the cystic artery by the inflammatory process is thought to be the main explanation of CAP rare incidence despite of the high prevalence of cholecystitis [7].

Most of the reported cases are discovered after CAP rupture which usually occurs into the GB and biliary tract causing hemobilia presented clinically with Quincke's triad, i.e., biliary colic, obstructive jaundice, and upper gastrointestinal tract (GIT) bleeding. However, this classic triad is only reported in less than 50% of cases [8]. Blood clots within the gallbladder or CBD may mimic stones on MRCP (Figure 4) or ultrasound, as occurred in our patient [9]. Unruptured CAPs may also be incidentally

detected on CT in the setting of acute cholecystitis [10]. Non-traumatic CAP has been reported as a rare cause of hemobilia [11]. Bleeding severity varies widely, from mild anemia to massive hemorrhage causing hypovolemic shock [4]. Fresh bleeding per rectum is a very rare presentation of CAP rupture in cases with cholecystocolonic fistula [12]. Cystic artery pseudoaneurysm may rarely rupture into the peritoneal cavity if GB is perforated causing hemoperitoneum [13].

In cholecystitis-related CAP, cholecystectomy with pseudoaneurysm resection was reported as urgent treatment in about 39% of reported cases and elective treatment after pseudoaneurysm embolization in 34.8% of cases while CAP treatment with embolization only was reported in about 26% of cases. Most reports consider transarterial embolization as a temporary treatment to stabilize the patient and reduce risk of pseudoaneurysm rupture during cholecystectomy [1].

Coils are the most used material for embolization in the reported cases (about 68.5% of cases) however pseudoaneurysm refilling can occur if coils are not densely packed within the aneurysm or if feeding artery was coiled proximally with increased risk of distal refilling of the pseudoaneurysm from collateral vessels [14]. Coils were used in conjunction with other embolizing agents like gel foam, NBCA, and thrombin in few reports (about 15% of reported cases). Other embolic materials were used as the main embolizing agent in few reports (NBCA in 5.6 %, gel foam in 3.7%, polyvinyl alcohol (PVA) in 1.85% and thrombin in 3.7% of reported cases). If the neck of the pseudoaneurysm is wide, then covered stent insertion could be an option to occlude the pseudoaneurysm neck while preserving patency of hepatic arteries. However, embolization of the right hepatic artery is reported to be relatively safe and the risk of hepatic necrosis is extremely low because of the presence of intrahepatic collaterals between right and left hepatic arteries and patency of the portal vein [15]. In our case, NBCA was used to embolize the whole pseudoaneurysm and cystic artery stump instead of using coils because of the large size of the pseudoaneurysm necessitating the use of many coils and the need for faster embolizing material due to the critical condition of the patient.

Although theoretically expected, gallbladder ischemia following cystic artery embolization is rarely reported, likely due to collateral supply from the epicholedochal plexus [6]. The use of particulate agents may increase the risk of end-organ ischemia [16]. In our patient, severe pre-existing inflammation combined with NBCA use likely contributed to gallbladder ischemia and subsequent perforation.

Percutaneous cholecystostomy is considered as an effective treatment for resolving acute cholecystitis and safe management option in critically and surgically unfit patients [17]. There are two reported cases of CAP and perforated GB in surgically unfit patients successfully treated by transarterial embolization for CAP and percutaneous cholecystostomy for perforated GB [2, 18].

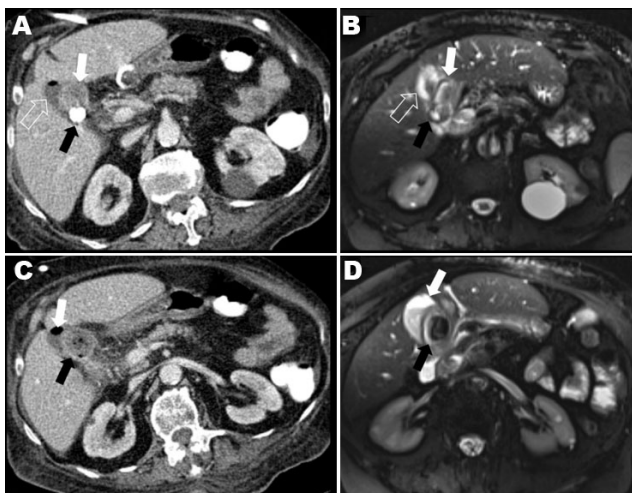


Figure 4: Correlation of post embolization CT and pre-embolization MRI. (A) and (B) Axial CT and MRI images at the same level showing the pseudoaneurysm (black solid arrow), the surrounding hematoma (white solid arrow), and the residual GB lumen (hollow arrow). (C) and (D) Axial CT and MRI images at the same level showing liquified hematoma (black solid arrow) and residual GB lumen (white solid arrow).

Similarly, percutaneous drainage of the perforated GB was done successfully in our surgically unfit patient raising the concern about whether cholecystectomy is required after successful embolization of cystic artery pseudoaneurysm or not in those high-risk patients for surgery.

## CONCLUSION

Gallbladder ischemia and perforation are rare but important complications of cystic artery embolization, particularly when NBCA is used in the setting of severe inflammation. Early recognition and minimally invasive percutaneous drainage can prevent morbidity and provide a safe alternative to surgery in high-risk patients.

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## Acknowledgments

The authors acknowledge Microsoft Copilot software for assisting with language editing of the manuscript.

## Artificial intelligence (AI) use in the article:

Microsoft Copilot was used to assist with language editing of the manuscript. The tool was not used to generate any data in the article. All AI-generated suggestions were reviewed and approved by the authors. All content remains the authors' original work, and the authors take full responsibility for the final manuscript.

## Author Contributions

Ramy Ahmed – Conception of the work, Design of the work, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Abdelrahman Omar – Conception of the work, Acquisition of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

**Guarantor of Submission**

The corresponding author is the guarantor of submission.

**Source of Support**

None.

**Consent Statement**

Written informed consent was obtained from the patient for publication of this article.

**Conflict of Interest**

Authors declare no conflict of interest.

**Data Availability**

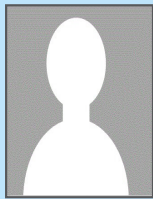
All relevant data are within the paper and its Supporting Information files.

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**Article citation:** Ahmed R, Ahmed A. Perforated gallbladder after cystic artery pseudoaneurysm embolization: A case report and review of the literature. *J Case Rep Images Surg* 2026;12(1):20–24.



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