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A challenging case of hemobilia: Endovascular treatment of cystic artery pseudoaneursym secondary to acute cholecystitis

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Abstract

Acute cholecystitis is a leading cause of individuals seeking medical attention in the emergency department due to right upper quadrant pain. Common complications associated with this clinical condition, which is frequently encountered in daily practice, include gangrenous cholecystitis, pericholecystic abscess, cholangitis, and gallbladder perforation. It is important to also consider the rare but potentially severe complication of cystic artery pseudoaneurysm. This case report details the development of a cystic artery pseudoaneurysm secondary to acute cholecystitis and its subsequent endovascular treatment in a 59-year-old male patient with no known underlying medical conditions, in accordance with existing literature.

Keywords: cystic artery, pseudoaneurysm, hemobilia

Introduction

Cystic artery pseudoaneurysm is an exceedingly uncommon complication, typically arising as a consequence of biliary system interventions but occasionally observed, albeit rarely, following acute cholecystitis [1]. In instances of complicated cases, inflammatory alterations within the pericholecystic region and potential concomitant hemorrhage may pose challenges for accurate detection through imaging modalities. Failure to promptly diagnose and treat this condition can result in significant morbidity and mortality.

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Informed Consent
The authors stated that the written consent was obtained from the patient presented with images in the study.

Conflict of Interest
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Case presentation

A 59-year-old male patient, previously free of known medical conditions, presented to our hospital's emergency department with complaints of progressively worsening right upper quadrant pain, accompanied by nausea and vomiting over the past two weeks. Upon physical examination, positive findings included defense and rebound tenderness, while the patient exhibited a body temperature of 38°C. Laboratory tests revealed elevated hepato-pancreaticobiliary enzyme levels, specifically: aspartate aminotransferase at 123 IU/L (normal range: <35), alanine aminotransferase at 197 IU/L (normal range: <50), alkaline phosphatase at 651 IU/L (normal range: < 119), γ-glutamyltransferase at 1275 IU/L (normal range: <73), total bilirubin at 2.7 mg/dL (normal range: <1.2 mg/dL), and direct bilirubin at 2.2 mg/dL (normal range: <0.3 mg/dL). The patient's leukocyte count was elevated at 14.58×10^3 cells/mL, while other laboratory parameters remained within normal limits.

An ultrasound examination was conducted to assess the patient, given the preliminary diagnosis of acute cholecystitis, revealing an enlarged gallbladder (transverse diameter: 4 cm). The gallbladder contained concentrated bile sludge and a few stones with a diameter of 1.5 cm. These findings were consistent with acute cholecystitis. Subsequently, a contrast-enhanced upper abdomen computed tomography (CT) scan was performed to provide a comprehensive evaluation and identify potential additional pathologies. The CT scan revealed hydropic changes in the gallbladder, marked thickening and irregularity in the gallbladder wall, and stranding in the pericholecystic area. Furthermore, dilation of the intrahepatic bile ducts and a common bile duct diameter of 16 mm were observed. Density increments suggestive of possible stones or biliary sludge were identified in the lumen distal to the common bile duct. The pancreas appeared normal in the imaging (Figures 1 and 2).

The patient, who was admitted with acute cholecystitis, was initiated on treatment protocols involving Ceftriaxone (2 × 1 gr) and IV Metronidazole (4 × 500 mg) upon hospitalization. Subsequently, the patient underwent endoscopic retrograde cholangiopancreatography for choledocholithiasis, during which a biliary tract stone was successfully removed, and a 10F stent was placed due to the blunt distal end of the common bile duct. Additionally, brush cytology was performed from the distal common bile duct at the conclusion of the procedure, revealing findings consistent with hemorrhage.

A follow-up hepatobiliary ultrasound examination of the patient detected an echogenic fluid collection extending up to 6 cm in the gallbladder fossa, primarily suggestive of hemobilia. To further elucidate the etiology, a subsequent dynamic upper abdomen CT scan was performed. The gallbladder walls displayed irregularity, making it challenging to assess wall integrity. Within the gallbladder lumen, a prominently opacified 9 mm diameter pseudoaneurysm was observed, particularly during the arterial phase. Furthermore, heterogeneous density increases were noted in the surrounding adipose tissue in the subhepatic area and the paracolic area adjacent to the gallbladder. The CT examination confirmed the large-sized hemobilia previously identified in the ultrasound (Figure 3).

Figure 1: Upper abdomen CT revealed hydropic and heterogeneous gallbladder and mucosal hyperenhancement which resembled acute cholecystitis (arrow), pancreas head was normal (star)

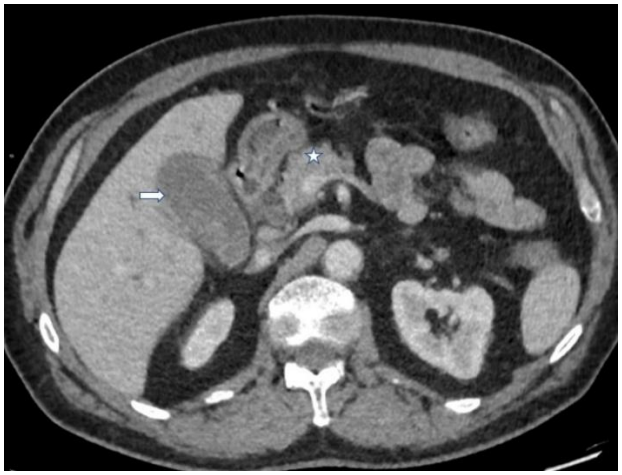
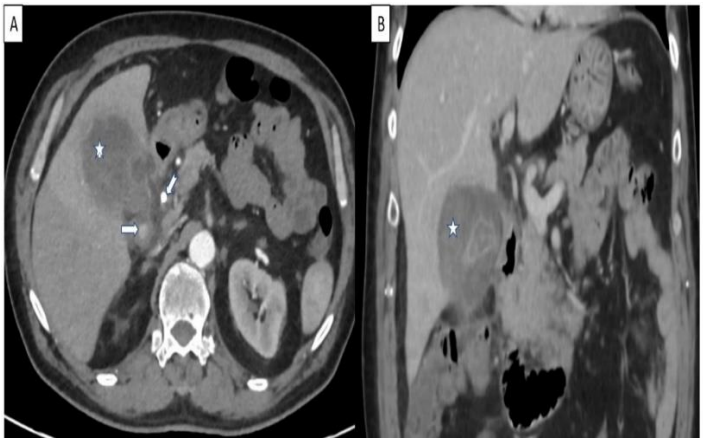


Figure 2: Upper abdomen CT showed a nodular lesion in the portal venous phase. The lesion was thought to be a gallbladder stone. The following imaging modalities revealed that was a pseudoaneursym (arrow)

Figure 3: A.B Dynamic upper abdomen CT revealed cystic artery pseudoaneurysm in the arterial phase (arrow) and giant hemobilia at the gallbladder fossa (star), biliary stent (notched arrow)



Following these findings, the patient was referred to the interventional radiology department. Superselective angiography revealed the origin of the pseudoaneurysm from a branch of the cystic artery. The cystic artery was successfully embolized using coils, resulting in the disappearance of the aneurysm during follow-up (Figures 4 and 5). No complications were observed throughout the monitoring period, and the patient was discharged with appropriate recommendations.

Figure 4: A.B Super selective angiography revealed cystic artery pseudoaneurysm (arrow), the pseudoaneursym was embolized with coils and the aneurysm disappeared in the control (notched arrow)

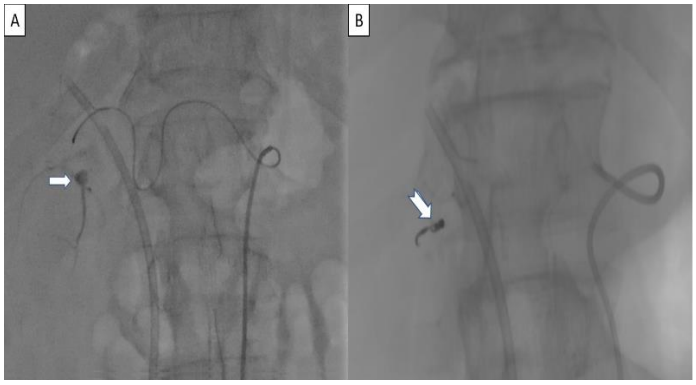
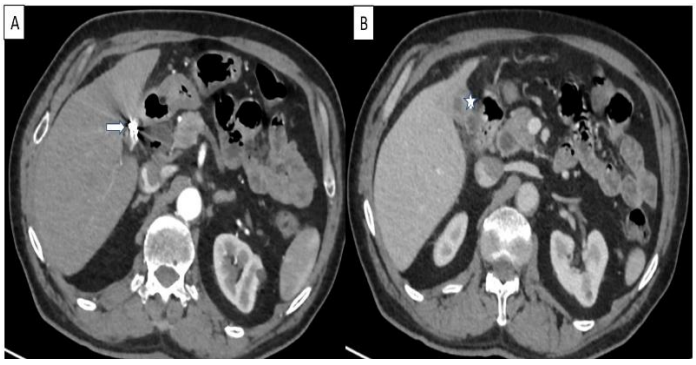


Figure 5: A.B After the six months of embolization, control upper abdomen CT revealed embolization material in the gallbladder fossa (arrow), post-operative changes in the gallbladder (star)



Discussion

Acute cholecystitis stands out as one of the most prevalent causes of right upper quadrant pain encountered in daily medical practice. Patients typically present with symptoms such as fever, nausea, vomiting, and right upper quadrant pain. If acute cholecystitis is inadequately treated, it can lead to severe complications, including pericholecystic abscess, cholangitis, necrosis in the gallbladder, gallbladder perforation, and peritonitis [2,3]. Another noteworthy complication in untreated cholecystitis cases in the chronic phase is the development of a cholecystoduodenal fistula [4].

While acute cholecystitis is relatively common, the occurrence of a pseudoaneurysm in the context of acute cholecystitis is exceedingly rare and is mainly documented in isolated case reports in the literature [5,6]. The most prevalent predisposing factor for the development of cystic artery pseudoaneurysms is a history of previous cholecystectomy surgery [7]. It has been suggested that inflammation secondary to acute cholecystitis may weaken the arterial wall, serving as the underlying etiology for this condition [8]. Patients with cystic artery pseudoaneurysms typically present with symptoms like hemobilia, hemoperitoneum, and occasionally upper gastrointestinal bleeding. Clinical manifestations, often referred to as Quincke's triad, include upper abdominal pain, obstructive jaundice, and gastrointestinal hemorrhage [9].

Although a characteristic yin-yang pattern is described in Doppler ultrasound for detecting cystic artery pseudoaneurysms, ultrasound alone may present challenges in diagnosis. In cases where rupture occurs, hyperechoic fluid may be observed in the gallbladder lumen on ultrasound, warranting consideration of an underlying pseudoaneurysm. CT angiography is deemed the most effective diagnostic tool for evaluating major arteries in the context of intra-abdominal inflammatory conditions. It can reveal anatomical variations and detect pseudoaneurysms. In our specific case, the initial CT scan lacked precontrast images, leading to a misinterpretation of the cystic artery pseudoaneurysm as a stone. However, upon reevaluation with a dynamic CT scan that included precontrast and arterial phase images, the true nature of the lesion as a cystic artery pseudoaneurysm became evident. In certain instances, the diagnosis of small-sized pseudoaneurysms may necessitate standard catheter angiography [10].

The management of cystic artery pseudoaneurysms remains a subject of debate in the medical community. While there have been reports of successful non-operative management of hemobilia, the majority of recommendations lean towards interventional approaches, such as selective visceral angiography with embolization or surgical intervention [11]. Furthermore, percutaneous selective cystic artery embolization has emerged as an effective treatment strategy, boasting lower mortality and morbidity rates, improved identification of the bleeding vessel, and enhanced hemorrhage control compared to surgical options. A significant advantage of this approach is its ability to combine both diagnosis and treatment within the same procedure session [12].

Moreover, some publications advocate for performing an elective cholecystectomy following embolization in patients with cystic artery pseudoaneurysm [13]. However, it is worth noting that in the case of our patient, they declined the cholecystectomy procedure, making it impossible to proceed with that aspect of the treatment.

The embolization process can employ various materials such as coils, N-Butyl cyanoacrylate (glue injection), or Gelfoam. Coil embolization stands as the most commonly employed method, with different coil sizes were chosen based on the vessel's diameter during the procedure [14]. It is important to note that some other embolizing agents carry the risk of potentially elevating pressure within the vascular lesion, which may lead to rebleeding after endovascular embolization [15].

Conclusion

While the occurrence of a cystic artery pseudoaneurysm in the context of acute cholecystitis is rare, it should remain a noteworthy consideration as a significant cause of hemobilia when formulating a differential diagnosis.

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