Case Report

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An unusual case of acute cholecystitis complicated by haemobilia and Mirizzi-like obstruction: a case report and review of literature

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Introduction: Cystic artery pseudoaneurysm rupture presents a rare yet potentially fatal aetiology for upper gastrointestinal (GI) bleed. While uncommon, its incidence has been rising with increased hepatobiliary surgical interventions, predominantly attributed to iatrogenic injury and rarely secondary to acute cholecystitis. Clinical manifestations typically include epigastric pain, upper GI haemorrhage, and obstructive jaundice. Due to its rarity, it is often excluded from initial differential diagnoses.

Case report: This is an unusual case of a 54-year-old male who presented with acute cholecystitis complicated by haemobilia and Mirizzi-like obstruction, in the setting of cystic artery pseudoaneurysm rupture. Initially, urgent transcatheter angiographic embolization of the cystic artery was performed to achieve hemodynamic stability. However, a triphasic computed tomography (CT) scan revealed the first attempt was unsuccessful, necessitating a second embolization. Subsequent imaging confirmed satisfactory embolization; however, a small area of liver necrosis was observed adjacent to the gallbladder. The patient was stable at discharge from the hospital and had an uncomplicated interval cholecystectomy.

Discussion: This case highlights the complexity and challenges associated with diagnosing and managing cystic artery pseudoaneurysm rupture. Diagnosis often relies on arterial phase contrast-enhanced CT scan. While no guideline currently exist, management typically involves achieving hemodynamic stability through Transcatheter angiographic embolization, followed by interval cholecystectomy.

Conclusion: Early recognition and intervention are crucial in managing cystic artery pseudoaneurysm rupture to prevent life-threatening haemorrhagic shock. Clinicians need to consider this rare condition in patients with upper GI bleeding and abnormal liver function tests.

Keywords: acute cholecystitis, cystic artery pseudoaneurysm rupture, cystic artery pseudoaneurysm, gallbladder perforation, haemobilia, Mirrizi-like obstruction

Introduction

The gallbladder receives its primary blood supply from the cystic artery, located within Calot's triangle. In ~89% of individuals, this artery originates from the right hepatic artery. However, variations may occur, with the cystic artery arising from the gastroduodenal artery, left hepatic artery, or superior mesenteric artery instead[1–3]. Pseudoaneurysm of the cystic artery is a rare condition that can lead to life-threatening haemorrhage if ruptured. This condition can occur post-cholecystectomy, idiopathically, or as a result of cholecystitis[4]. The exact pathophysiology is not fully understood; however, it is thought to be a consequence of inflammation in the arterial wall. This inflammation can cause erosion to both its elastic and muscular components, ultimately damaging the adventitia and weakening the vessel wall, leading to the formation of a pseudoaneurysm. Another theory suggests that early thrombosis of the cystic artery due to inflammation may also play a role in contributing to pseudoaneurysm formation[2,5].

In most cases, cystic artery pseudoaneurysm presents with abdominal pain (77.9%), gastrointestinal (GI) haemorrhage

HIGHLIGHTS

- Cystic artery pseudoaneurysm rupture is a condition of rare occurrence.
- Prompt recognition and early intervention are necessary to avoid unfavourable outcomes.
- There are no guidelines currently on the management of cystic artery pseudoaneurysm. However, it is often managed by cystic artery angioembolization followed by interval cholecystectomy.
- Liver necrosis is a rare complication of cystic artery angioembolization.

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(64.4%), and Quinke's triad (epigastric pain, GI haemorrhage, and obstructive jaundice) (32.2%)[2,6]. The presentation of haemobilia can vary depending on its duration and severity, including symptoms and signs of anaemia, obstructive clots, or even haemorrhagic shock[1]. Mirizzi syndrome is a rare condition characterised by compression of the common bile duct or common hepatic duct, typically caused by a gallstone[7]. In some instances, like our patient's case, obstruction may result from fluid accumulation in the infundibulum, compressing the common bile duct. This condition is known as Mirizzi-like obstruction.

Cystic artery pseudoaneurysm is an extremely rare condition; a study that reviewed reported cases between 1991 and 2020 found only 67 cases, with just 51 of them associated with cholecystitis[4]. The combination of haemobilia in the setting of acute cholecystitis, cystic artery pseudoaneurysm rupture, and Mirizzi-like obstruction is extremely rare; to our knowledge, there might be only a few similar reported cases in the literature. This work has been reported in line with the SCARE 2023 criteria[8].

Case

A 54-year-old man presented to the hospital via ambulance with acute epigastric pain and abnormal liver function tests. Three weeks prior to this admission, he had been hospitalised for a similar presentation, during which he was diagnosed with cholecystitis due to a stone in the neck of the gallbladder, as confirmed by ultrasound. He received conservative management with antibiotics and analgesia, then was scheduled for interval cholecystectomy before being discharged home.

On this admission, his blood test results were:

Total white cell count (TWCC): 5.8×10^9/L (normal: 4–10)
Haemoglobin (Hb): 11.4 g/dl (normal: 13.0–17.0)
C-reactive protein (CRP): 8.6 mg/l (normal: 0–5)
Total bilirubin: 30 umol/l (normal: 1–21)
Alkaline phosphatase (ALP): 593 U/l (normal: 40–129)
Alanine transaminase (ALT): 70 U/l (normal: 0–40)
Gamma-glutamyl transpeptidase (GGT): 612 U/l (normal: 10–71)

Ultrasound (US) scan revealed persistent cholecystitis with an impacted stone in the gallbladder neck and dilated common bile duct. Subsequently, magnetic resonance cholangiopancreatography (MRCP) showed an increase in conspicuity and severity of the cholecystitis, now accompanied by a fluid collection in the region of the infundibulum and cystic duct, suggestive of a localised perforation. This perforation caused extrinsic compression of the adjacent common hepatic duct, leading to Mirizzi-like obstruction (Fig. 1).

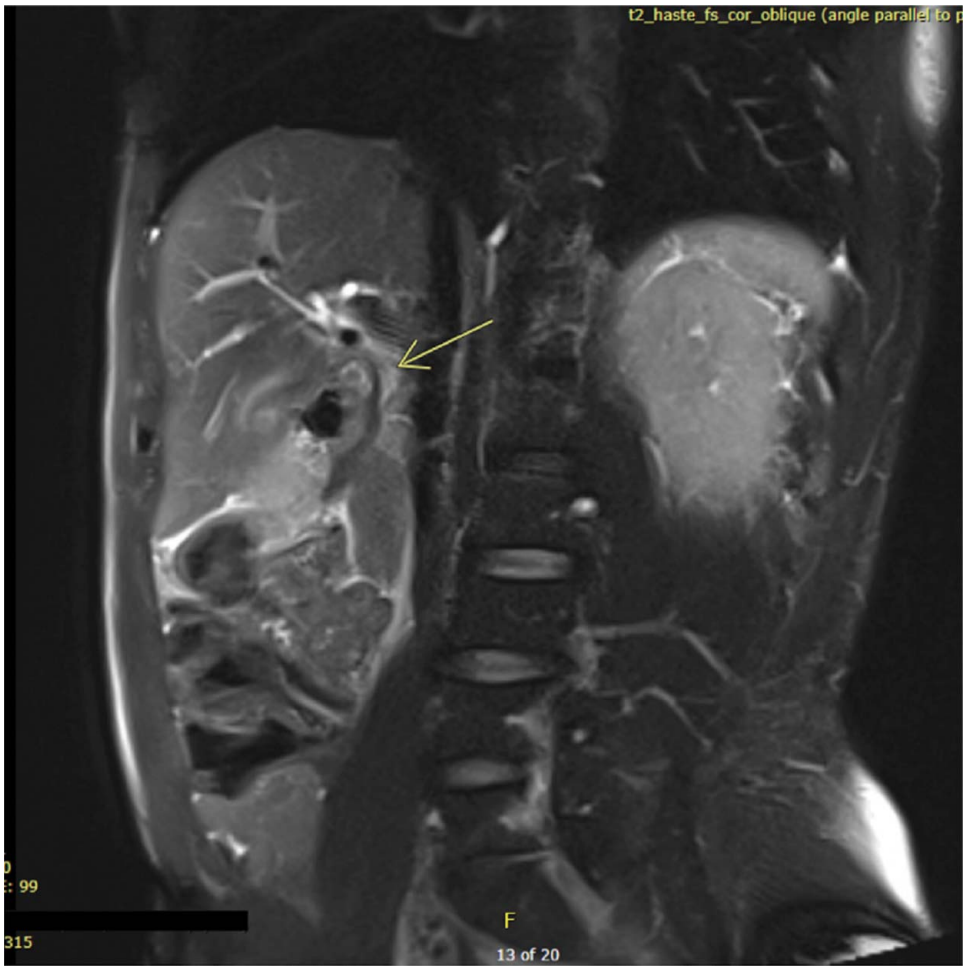

Figure 1. Magnetic resonance cholangiopancreatography showing a fluid collection in the region of the infundibulum and cystic duct, indicative of a localised perforation. The fluid collection is causing an extrinsic compression of the adjacent common hepatic duct, resulting in Mirizzi-like obstruction.

The following day, the patient had coffee-ground vomiting, prompting an oesophagogastroduodenoscopy. The oesophago-gastro-duodenoscopy (OGD) revealed mild nodular duodenitis with evidence of bleeding and erosion observed at the junction between D1 and D2 (Fig. 2). One day later, the patient exhibited diaphoresis, severe pain, and restlessness. He vomited ~200 ml of fresh blood with clots and stomach contents. An endoscopic retrograde cholangiopancreatography (ERCP) was then performed, which detected fresh blood in the stomach and D1, along with a large blood clot emerging from the ampulla. To relieve the obstruction, a biliary stent was inserted during the ERCP (Fig. 3). As a result of bleeding, his haemoglobin dropped to 6.1 g/dl, necessitating the transfusion of 2 units of blood.

CT-angiography showed active bleeding into the common bile duct, most likely originating from a branch of the cystic artery (Fig. 4). Subsequently, a Transcatheter angiographic embolization of the cystic artery was performed. The procedure involved careful access to the right common femoral artery, selective cannulation of the coeliac trunk, and embolisation of the cystic artery using gelfoam slurry, then coil embolization using a 4 mm × 5 cm and a 5 mm × 5 cm pushable coil (Fig. 5).

Two days later, a triphasic-computed tomography (CT) scan of the liver showed active contrast extravasation from the cystic artery, and the coil was noted to be in a branch of the right hepatic artery instead of the intended cystic artery, indicating an unsuccessful first attempt. Repeat angio-embolization of the cystic artery performed, which involved challenging selective catheterization due to complex anatomy. Approximately 10 microcoils (4 × 2 mm) were placed in the cystic artery to achieve complete vascular occlusion (Fig. 6). Completion angiography showed no flow to the cystic artery, and the patient returned to HDU in a stable condition.

Finally, the patient had a repeat Triphasic-CT scan 1 week post-embolization, showing satisfactory obliteration of the cystic artery pseudoaneurysm, with a small area of liver necrosis adjacent to the gallbladder (Fig. 7). Consequently, he was discharged and scheduled for an interval laparoscopic cholecystectomy. Our team conducted a three-month follow-up via phone calls, during which the patient reported an uncomplicated laparoscopic cholecystectomy performed two months after angio-embolization at a specialised hepatobiliary centre, without any preoperative or postoperative complications.

Discussion

Pseudoaneurysm of the cystic artery is a rare condition often characterised by abdominal pain, gastrointestinal haemorrhage,

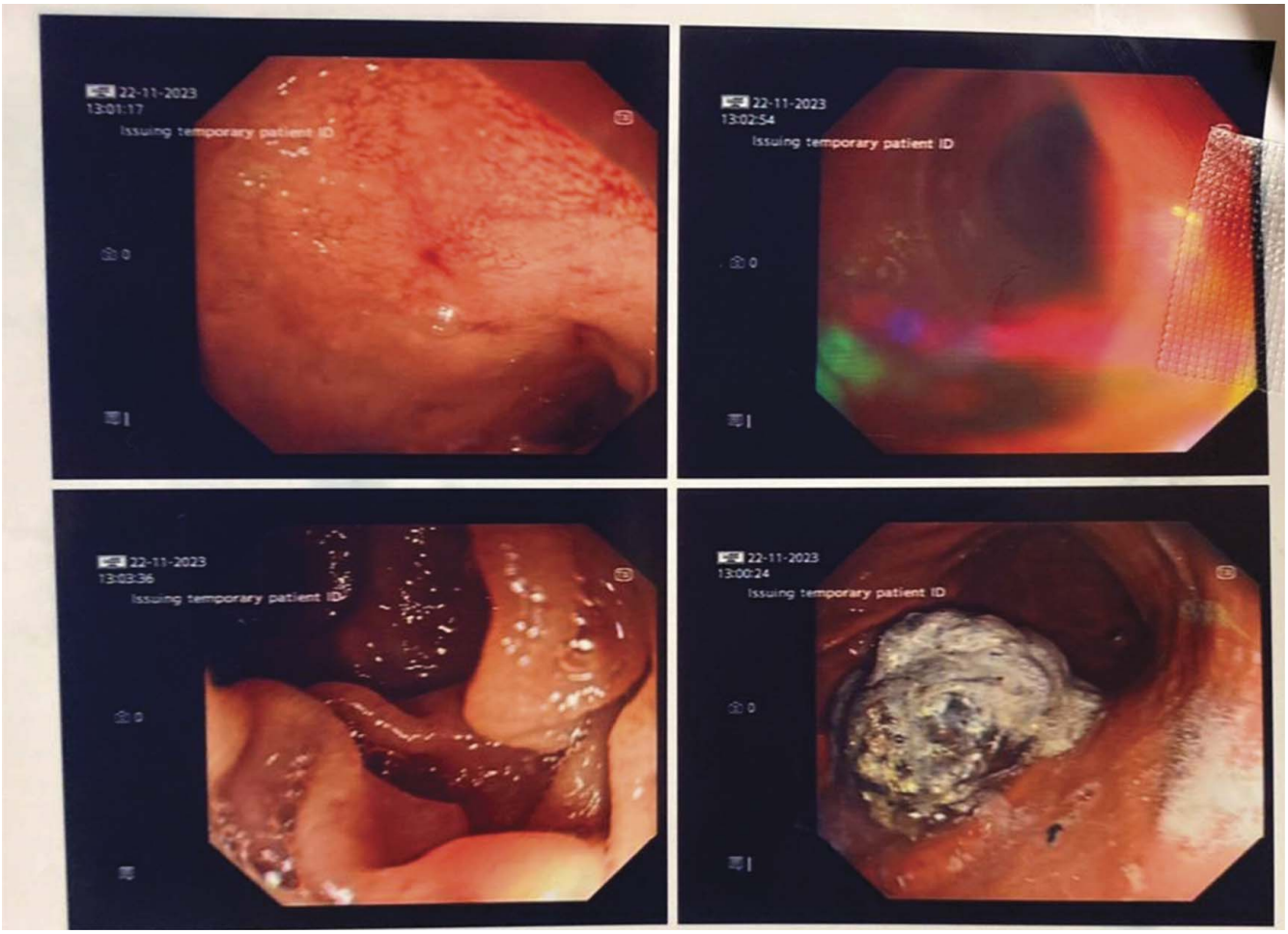

Figure 2. Oesophago-gastro-duodenoscopy showing mild nodular duodenitis with some clots, and erosion seen at the junction between D1 and D2.

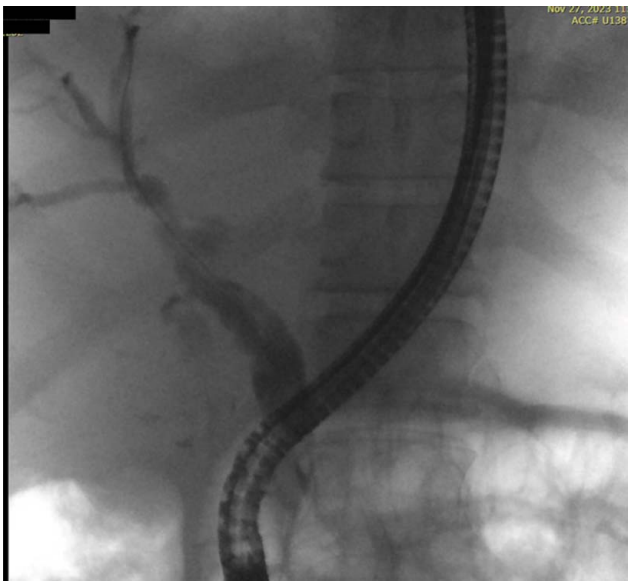

Figure 3. Endoscopic retrograde cholangiopancreatography was carried out on the patient and a stent was placed.

and Quinke's triad[2,5]. If left untreated, a ruptured cystic artery pseudoaneurysm can lead to fatal haemorrhagic shock[1]. The most commonly utilised imaging for detecting haemobilia is an arterial phase contrast-enhanced CT scan. OGD can detect clots and active haemorrhage in D2 only in 60% of cases, and ERCP may detect haemobilia but cannot locate the source of bleeding[4].

Currently, there are no established guidelines for the management of ruptured cystic artery pseudoaneurysm (CAP); however, it is frequently managed by transcatheter angiographic embolization to restore hemodynamic stability[9]. This approach is preferred over emergency cholecystectomy, which can present challenges for surgeons who must manage the haemorrhage while navigating an inflamed gallbladder. Nevertheless, transcatheter angiographic embolization carries a 2% risk of complications, including hepatobiliary necrosis, bleeding, abscess formation, catheter-related and contrast-related complications[2,4]. Moreover, access to interventional radiologists may be limited in some hospitals, particularly outside regular working hours. Following transcatheter angioembolization (TCA), interval cholecystectomy is recommended to prevent recurrent haemobilia and gallbladder ischaemia[10].

There is limited research comparing delayed versus emergency cholecystectomy after cystic artery pseudoaneurysm embolization. However, delayed cholecystectomy generally allows for

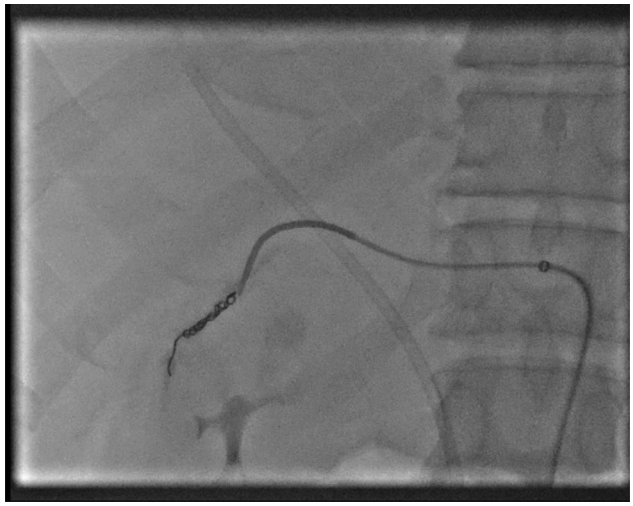

Figure 5. Transcatheter angiographic embolization of the cystic artery using a 4 mm × 5 cm and a 5 mm × 5 cm pushable coil.

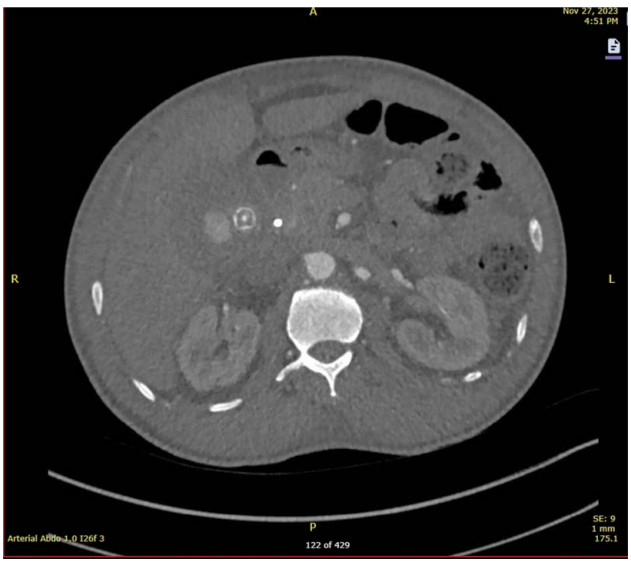

Figure 4. Computed tomography-angiography showing active bleeding into the common bile duct, most likely originating from a branch of the cystic artery.

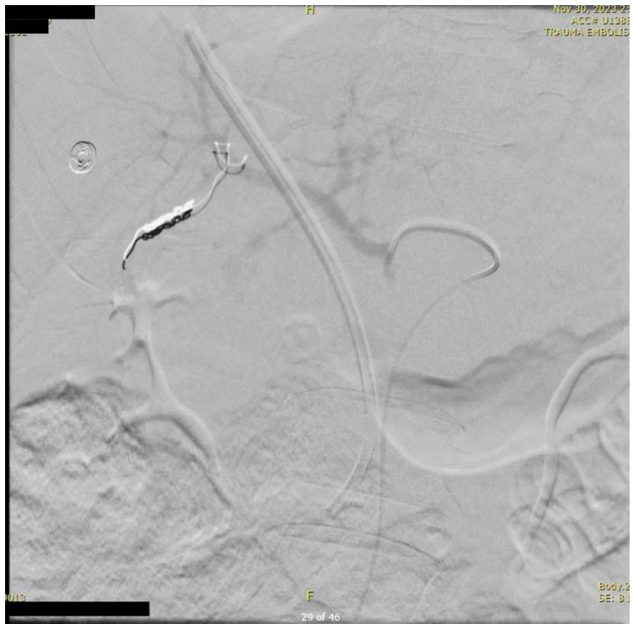

Figure 6. A second attempt at cystic artery embolization using 10 X (4 × 2 mm) diameter microcoils.

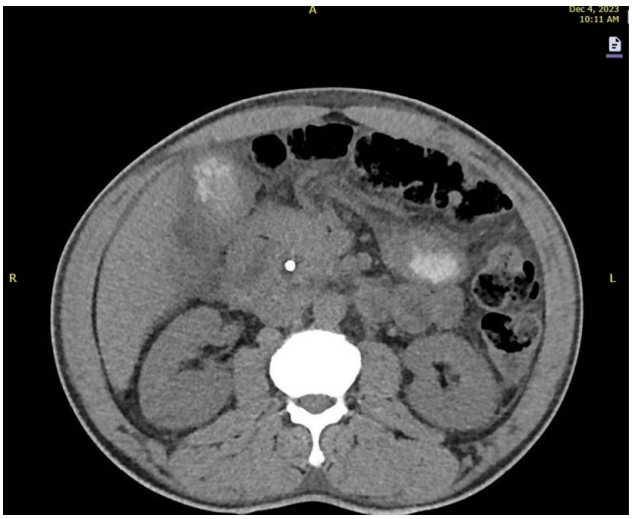

Figure 7. Repeat triphasic-computed tomography 1 week post-embolization, showing satisfactory obliteration of the cystic artery pseudoaneurysm, with a small area of necrosis in the liver adjacent to the gallbladder.

inflammation resolution, leading to a more favourable surgical environment and preoperative patient stabilisation. Additionally, it is linked to a reduced risk of conversion to open cholecystectomy, decreased intraoperative blood loss, and shorter postoperative hospital stays[11]. Upon reviewing the literature and comparing cases involving cholecystectomy and cystic artery ligation with those of transcatheter angiographic embolization, we observed similar outcomes between the two approaches, with no reported post-interventional complications[12–20]. However, recovery time after intervention was often unreported, necessitating further research to evaluate intervention efficacy of each intervention.

While managing this patient, we encountered several challenges. Firstly, there were frequent delays in receiving scan results, leading to delayed diagnosis. Secondly, the rarity of the condition initially excluded it from consideration in our differential diagnosis, potentially delaying its recognition. Thirdly, the case escalated to an emergency status late at night, presenting time-sensitive challenges that likely impacted the overall outcome. Lastly, the patient had a complex anatomy of his hepatobiliary arterial system, further complicating embolization and resulting in an unsuccessful initial attempt.

A less frequently reported side effect associated with TCA is hepatobiliary ischaemia. The risk of this complication remains low as TCA primarily targets the cystic artery for embolization. However, follow-up triphasic CT scans of our patient revealed a small area of liver necrosis adjacent to the gallbladder, possibly stemming from occlusion of the proximal common hepatic artery following partial dissection during the initial embolization attempt.

In summary, the patient was successfully managed with TCA, and hemodynamic stability was achieved. The only complication noted was a small area of liver necrosis. He was discharged two weeks following the second embolization and underwent an elective laparoscopic cholecystectomy two months after discharge, with no reported complications.

Conclusion

This case emphasises the importance for clinicians to be vigilant about haemobilia arising from cystic artery pseudoaneurysm rupture in patients presenting with upper GI bleeding and abnormal liver function tests. Given its rarity and complexity, identifying this condition can pose significant challenges. Early recognition and intervention are crucial for favourable outcomes, as untreated ruptured CAP can lead to potentially life-threatening haemorrhagic shock.

Ethical approval

This case report exclusively focuses on an individual patient, and we have obtained explicit consent for the disclosure of their medical information and imaging. As our submission is solely a case report, without involving experimental interventions or research procedures, formal ethical approval from an ethics committee is not applicable in this context. The manuscript adheres rigorously to patient confidentiality standards, ensuring the exclusion of any identifiable information that could compromise the individual's privacy. Please do not hesitate to reach out for further clarification or additional information.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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None.

Author contribution

A.K.: wrote, collected data, and edited the case report. M.M.K.: proofread the case report, provided input on the case and editorial advice. Helped in finding the relevant figures. R.G.: Supervising consultant.

Conflicts of interest disclosure

Not applicable.

Research registration unique identifying number (UIN)

Our study is a case report.

Guarantor

Ahmed Khawjah, Mugahid M Khair and R. Goubran.

Data availability statement

Not applicable.

Provenance and peer review

No peer review has been conducted yet.

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