Post-laparoscopic Cholecystectomy Cystic Artery Stump Pseudoaneurysm Presenting With Haemobilia and Hematochezia: a Case Report With Literature Review

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Abstract

**Background:** Cystic artery stump pseudoaneurysm (CASP) is a potentially life-threatening condition that can be related to multiple etiologies, especially the iatrogenic factor owing to the increased number of hepatobiliary procedures. Most patients present with haemobilia. Here we report a rare case of unusual presentation of CASP with recurrent haemobilia and hematochezia.

**Case presentation:** A 38-year-old patient developed bile duct injury after laparoscopic cholecystectomy which was identified by magnetic resonance cholangiopancreatography (MRCP). Later, she developed haemobilia due to CASP which was then treated by trans-arterial embolization (TAE) followed by fresh bleeding per rectum one month later. The latter complication eventually required surgical ligation.

**Conclusions:** CASP is a rare complication after cholecystectomy that can present as upper gastrointestinal bleeding or rarely as fresh bleeding per rectum.

**Level of evidence:** Level 4, Case report and literature review

**Background:**

A Pseudoaneurysm is defined as an abnormal outpouching or dilatation of an artery which is bounded only by the outer most layer of the arterial wall (tunica adventitia), compared to a true aneurysm that is bound by all three layers of the arterial wall [1]. Pseudoaneurysm formation is a consequence of vascular injury caused by trauma, or more commonly iatrogenic causes, including hepatobiliary biopsies and surgeries [2]. We are reporting a case of pseudoaneurysm in the cystic artery, which was diagnosed through computed tomography (CT) and angiographic evaluations and was successfully treated with surgical ligation after the failure of TAE.

**Case Presentation:**

A 38-year-old female with an unremarkable medical history, presented to the emergency department (ED), complaining of right upper quadrant (RUQ) pain, nausea and vomiting 11 days after a laparoscopic cholecystectomy (LC). There was no evidence of fever, hematemesis, or melena. Bedside physical examination was positive for RUQ tenderness. Basic laboratory investigations were unremarkable except for cholestatic liver enzyme elevation.

Ultrasound (US) scan of the abdomen showed a well-defined subhepatic collection within the gallbladder fossa and minimal abdominopelvic free fluid. Subsequently, MRCP showed T2 heterogenous, and T1 hypointense gallbladder fossa collection, with rim enhancement and no internal restricted diffusion, with a suspected communication between collection's upper part and the adjacent hepatic duct confluence, concerning for bile duct injury (Figure 1). Common hepatic duct and common bile duct were of normal caliber. Subsequent Endoscopic retrograde cholangiopancreatography (ERCP) confirmed bile duct injury and stricture, and accordingly, a plastic stent was placed across the injury (Figure 2). CT-guided drainage of the abdominal collection, was done by inserting 8.3 French drainage catheter in the right subhepatic collection, followed by aspiration of 50 ml of orange/yellow bile with debris.

Follow-up abdominal CT showed significant reduction in gall bladder fossa collection size, with complete resolution of the abdominopelvic ascites. However, a suspicious small contrast-filled outpouching was seen at the porta hepatitis, close to cholecystectomy clips, raising the possibility of a cystic artery stump pseudoaneurysm (CASP) (Figure 3a). Until then, the patient was clinically asymptomatic. Five days later, the patient developed itching, jaundice, melena and acute drop of hemoglobin of 3 mg/dl. Therefore, haemobilia was suspected. Subsequent abdominal CT angiogram (Figure 3b-d) demonstrated an increase in the size of the contrast filled porta hepatitis outpouching, closely related to the cholecystectomy clip, suggesting CASP. Hereafter, selective common hepatic artery angiogram confirmed the presence of CASP, which was coiled by six micro coils (Figure 3e-f). Thereafter, the patient recovered and was discharged home.

One month later, the patient presented to ED with massive bleeding per rectum (hematochezia), hypotension responding to fluid resuscitation, and drop of 4 mg/dl of hemoglobin. Correspondingly, esophagogastroduodenoscopy, colonoscopy, and conventional abdominal angiography were negative for bleeding. Bleeding from pseudoaneurysm was still suspected, and surgery was unavoidable in this case. Therefore, excision of the pseudoaneurysm and right hepatic artery repair at the site of the cystic artery origin with Roux-en-Y hepaticojejunostomy and adhesiolysis (due to significant adhesions found intraoperatively) was achieved. Patient recovery was uneventful, and follow-up was assuring.

**Conclusions:**

Hepatic artery or cystic artery pseudoaneurysms are rare complications of laparoscopic cholecystectomy, with cystic artery involvement being reported much less frequently in the literature. It is hard to determine the incidence of the pseudoaneurysm, as it could be asymptomatic, thrombosed or ruptured [2, 3]. The reported time interval between the surgery and onset of clinical symptoms is variable, with one month reported as the average time to presentation. However, a 5-year delay in presentation has been described [2].

The exact mechanisms of hepatic or cystic artery pseudoaneurysm after laparoscopic or open cholecystectomy is not completely identified up to date, however, they can be related to direct vascular injury (by applying surgical clips/ thermal injury), adjacent gallbladder fossa bile collection that increases vessel wall fragility or as a consequence of post-surgical adhesions. Most of the cases presented with classical symptoms of haemobilia (gastrointestinal bleeding, upper abdominal pain, and jaundice) [4], but it is rare to present with symptoms of lower GI bleeding [5].

Pseudoaneurysm is considered an acute emergency that requires immediate intervention. The diagnosis can be made by endoscopy, ERCP, CT and conventional angiography, and managed primarily by TAE through occluding the sac or the feeding vessel with a variety of embolic agents, including coils,
thrombin, or gel foam, before ideally embolizing the vessel distal and proximal to the pseudoaneurysm to prevent its collateral filling [4]. Yet, surgical resection of the pseudoaneurysm and ligation of the cystic artery (CA) stump or right hepatic artery (RHA) would be an alternate treatment option if TAE fails.

Up to the authors’ knowledge, in the last five years, 46 cases of porta hepatitis pseudoaneurysm were reported [2, 4, 6–24] (Table 1). 52.1% (24 cases) were males, age range from 39 – 82 years, with mean age of 56.3 years, and 47.9% (21 cases) were females, age range from 37- 88 years and mean age of 61.1 years. The most common involved vessel was RHA (37 cases, 80.4%), followed by CA (8 cases, 17.4%), while left hepatic artery involvement was reported in one case. 41 cases were managed through laparoscopic approach while 5 cases were done through open resection and time of presentation ranged from day 1 up to 26 months post-procedure (mean of 60.4 days). The most common documented presentation was haemobilia, as reported in 31 cases (78.3%). Furthermore, abdominal pain, sepsis, jaundice, and hypotension were encountered, with only one case in the last five years presented with hematochezia. In 12 cases, iatrogenic bile duct injury was considered, however, no case documented the presence of bile collection/biloma. 2 out of 46 cases (4.3%) experienced an increased pseudoaneurysm size, however, 4 cases confirmed recurrence of bleeding episode (8.7%). 41 cases (89.1%) were successfully managed by TAE, while 3 cases (6.5%) required surgical ligation, 2 cases (4.3%) treated by percutaneous direct puncture and one case (0.1%) managed conservatively. Favorable outcome was achieved in 97.8% of reported cases.

In this case, we are discussing an unusual presentation and recurrence of CASP bleed, in which, the three assumed pathological factors for cystic artery/ RHA pseudoaneurysm formation are present, including vascular injury by surgical clips, post LC bile collection and extensive post-surgical adhesions. In the related literature review, only one similar presentation of fresh bleeding PR secondary to CASP was identify. Furthermore, the source of hematochezia in our case was difficult to localize by different investigative modalities. Accordingly, surgical intervention was the definite treatment.
| Author/ Year of the article | Number of cases | Age/ Gender | Type of surgery | Time to presentation | presenting complaint | Bile duct injury | Vessel Involved | Increased aneurysms’ size | TX |
|-----------------------------|----------------|-------------|----------------|---------------------|---------------------|-----------------|----------------|-------------------------|----|
| Rossini M et al., 2019 [2]  | 1              | 66/M        | OC             | 28 Days             | RUQ pain, Hemobilia | No              | CA             | No                      | TAE/Coils |
| Machado et al, 2017 [4]    | 1              | 70/F        | LC             | 14 Days             | Abdominal pain      | No              | RHA            | No                      | TAE/Coils |
| Gachabayov M et al., 2017 [12] | 1            | 57/M        | LC             | 15 Days             | Hemobilia, jaundice, abdominal pain | No              | RHA            | Yes                     | TAE/Alcohol particles + Surgical ligation |
| Wen F et al., 2016 [23]    | 1              | 49 m/ 10 F, 4 M | LC        | 21 Days (m)        | Hemobilia, abdominal pain, jaundice | No              | RHA            | No                      | TAE/Coils |
| CreTu O M et al., 2017 [11] | 1              | 55          | LC             | 22 Months           | Haemobilia + upper abdominal pain | No              | RHA            | No                      | Surgical ligation |
| Ion D et al., 2016 [14]    | 1              | 58/M        | OC             | 27 Days             | Haemobilia+ anemia + hematochezia | No              | RHA            | No                      | TAE |
| To K et al, 2018 [20]      | 1              | 56/M        | LC             | 28 Days             | Haemobilia + RUQ pain | No              | CA             | No                      | TAE/Coils + stent |
| Abiko T et al., 2020 [6]   | 1              | 60/M        | LC             | 3 Days              | Haemobilia + RUQ pain | No              | CA             | No                      | TAE |
| Choudhary A et al., 2017 [10] | 1         | 42/F        | LC             | 26 Months           | RUQ pain+ hematemesis | No              | CA             | No                      | TAE/coil + stent/distal bulge |
| Villa-Gomez G et al., 2018 [22] | 1      | 39/M        | LC             | 35 Days             | Hematemesis+ cholangitis | Yes             | RHA            | No                      | TAE |
| Badillo R et al., 2017 [9] | 1              | 79/M        | LC             | 15 Months           | Haemobilia          | No              | CA             | No                      | PDP |
| Arata R et al., 2020 [8]   | 1              | 88/F        | LC             | 12 Days             | Haemobilia + Abdominal pain + Back pain | No              | CA             | No                      | conservative |
| Traa AC et al., 2020 [21]  | 1              | 76/F        | LC             | 1 Day               | Pain+ haemobilia    | No              | RHA            | No                      | TAE/Coil+ stent/coil |
| Tiwari A et al., 2017 [19] | 1              | 80/M        | LC             | 30 Days             | UGIB+ hematochezia+ shock | No              | LHA            | No                      | TAE |
| Yagihashi K et al., 2017 [24] | 1        | 55/M        | OC             | 7 Days              | Fever + abdominal pain | No              | RHA            | No                      | PDP |
| Rege S A et al., 2017 [17] | 1              | 40/F        | LC             | 55 Days             | Hematemesis + RUQ pain | No              | CA             | No                      | TAE/Coils |
| Alrajraji M et al., 2016 [7] | 1            | 41/F        | LC             | 8 Months            | Hematemesis + melena (haemobilia) | No              | RHA            | Yes                     | Surgical ligation |
| Gandhi R J et al., 2020 [13] | 13       | 52m. 3F, 10M | LC           | 9-30 Days (15.6days=m) | 3 hematemesis, 2 Malena, 4 sepsis + anemia. 4 hypotension | Yes (9 cases) | RHA | N/A                     | TAE(3Coils,4NBCA,6Coil+NBCA) |
| Kassem T W et al., 2017 [15] | 1           | 42F         | LC             | 6 Months            | Hematemesis         | No              | RHA            | No                      | TAE/Coils |
| Rosa C et al., 2018 [18]   | 1              | 52F         | OC             | 4 Months            | Hematemesis + Malena (haemobilia) | Yes             | RHA            | No                      | TAE/NBCA |
| Author/ Year of the article | Number of cases | Age/ Gender | Type of surgery | Time to presentation | presenting complaint | Bile duct iatrogenic injury | Vessel Involved | Increased aneurysms' size | TX |
|----------------------------|----------------|-------------|-----------------|----------------------|----------------------|---------------------------|----------------|---------------------------|----|
| Mahfooz F et al., 2020 [16] | 1              | 82M         | OC              | 2 Months             | Abdominal pain + Malena | Yes                       | CA             | No                        | TAE |

CA: Cystic artery, RHA: Right hepatic artery, LHA: Left hepatic artery, TAE: Trans-arterial embolization, NBCA: n-butyl-2-cyanoacrylate, PDP: percutaneous direct puncture, LC: laparoscopic cholecystectomy, OC: open cholecystectomy, M: Male, F: Female, m: Mean, RUQ: right upper quadrant pain

CASP is a rare complication of post LC, yet a potentially life threatening, with possible delayed complications occurring months to years after the surgery. We are presenting a rare case of CASP presented with melena and fresh bleeding per rectum, with presence of all possible etiopathogenesis, diagnosed by contrast CT and angiogram, and eventually treated with surgical ligation after failure of the initial TAE. Clinicians and radiologists should be aware of this important entity and its variable manifestations to facilitate early treatment.

**List Of Abbreviations:**

- Cystic artery stump pseudoaneurysm (CASP)
- Magnetic resonance cholangiopancreatography (MRCP)
- Trans-arterial embolization (TAE)
- Computed tomography (CT)
- Emergency department (ED)
- Right upper quadrant (RUQ)
- Laparoscopic cholecystectomy (LC)
- Ultrasound (US)
- Endoscopic retrograde cholangiopancreatography (ERCP)
- Cystic artery (CA)
- Right hepatic artery (RHA)

**Declarations:**

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**Competing interests**

The authors declare that they have no conflict of interest.

**Ethical approval and consent to participate**

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

An approval from the clinical research Clinical Imaging Research Committee (CIRC) in Hamad Medical Corporation was obtained prior to submission of this manuscript.

Written informed consent was obtained from the individual included in the study.

**Consent for publication**

Consent for publication was obtained for the patient included in the study. However, no identifiable parts of the patients are demonstrated in the article.

**Availability of data and material**

Not applicable.

**Authors' contributions**
Dr. Sushila Ladumor, Dr. Maysa Abdulhafeez, Dr. Amal Al-Rashid, and Dr. Aalaa Kambal analyzed and interpreted the patient imaging studies and helped in writing the relevant findings in the paper. Dr. Ali Barah and Dr. Omran Almkdad helped in analyzing the interventional radiology procedures details and outcomes. Dr. Dalal Sibira, Dr. Mohamed F. Ahmed and Dr. Loai Aker helped in collecting patient relevant information and in writing the literature review.

All authors contributed in the writing and proofreading process and approved the final manuscript.

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Figures
Figure 1
Coronal 3-D reconstruction of magnetic resonance cholangiopancreatography image showing heterogenous high T2 signal gallbladder fossa collection (arrow), with suspected communication between collection's upper part and adjacent hepatic duct confluence (arrow head), concerning for bile duct injury. Moderate ascites is noted.

Figure 2
Intraoperative radiographic images of Endoscopic retrograde cholangiopancreatography (ERCP) demonstrating contrast leak (white arrow), after injecting contrast through ampulla of Vater (a), concerning of bile duct injury, followed by stent placement (red arrow) (b).
Figure 3

CT abdomen axial image at first follow-up after drainage of abdominal collection (a) showing small contrast-filled structure at the porta hepatis (arrow) close to cholecystectomy clips, possibly cystic artery stump. Serial axial images of CT abdominal angiogram study representing arterial (b), venous (c) and delayed acquisition phases (d), respectively, showing focal contrast-filled outpouching (arrows) at the porta hepatis along the lateral aspect of right hepatic artery closely related to cholecystectomy clip and biliary stent, with increasing size compared to previous exam (a). The lesion is appearing isodense to aorta on arterial phase image (b), and venous phase (c) and washing-out on delayed scan (d), corresponding to a cystic artery stump pseudoaneurysm. Selective common hepatic artery angiogram (e-f) confirmed cystic artery stump pseudoaneurysm (white arrow), which was coiled by six micro coils (red arrows).