

CASE REPORT

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# Delayed appearance of hepatic artery pseudoaneurysm and an arterio-biliary fistula secondary to biliary leakage and bilioma formation in a liver trauma patient and its management through transarterial embolization

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## Abstract

**Background** Pseudoaneurysm and arterio-biliary fistulas are infrequent complications of surgery or iatrogenic trauma, accidental trauma, tumors, vascular malformations, infection, and inflammation of the biliary tract. In a liver trauma patient, they evolve immediately secondary to direct vascular injury and usually present with either drop in hemoglobin level or hemobilia due to bleeding into the peritoneal space or biliary system. In the present case, there were no clinical features of hemobilia or upper gastrointestinal bleeding. Moreover, the hepatic artery pseudoaneurysm and arterio-biliary fistula evolved after a latent period of a few weeks, secondary to the toxic effects of the bile on the vessel walls due to biloma formation or biliary leakage, which is exceptionally uncommon.

**Case presentation** This 19-year-old male presented with a severe liver injury complicated by forming a biloma, followed by the development of a pseudoaneurysm and an arterio-biliary fistula. The appropriate hepatic artery branch was successfully embolized using an endovascular approach.

**Conclusions** In a liver trauma patient, the latency period for developing a pseudoaneurysm and an arterio-biliary fistula can be from a few days to a few months due to direct vascular injury or toxic effects of the bile on the adjacent vessel walls secondary to bile leakage. Development of hepatic artery pseudoaneurysm and an arterio-biliary fistula after a latent period of a few weeks, secondary to the toxic effects of the bile on the vessel walls due to biloma formation or biliary leakage, is exceptionally uncommon. So, these liver trauma patients require close monitoring and follow-up for early detection and treatment of pseudoaneurysm and arterio-biliary fistula, especially when there are clinical or imaging features of bile leakage or biloma formation. Endovascular treatment of hepatic artery pseudoaneurysm and arterio-biliary fistulas is a safe and effective method.

**Keywords** Hepatic artery pseudoaneurysm, Trauma, Arterio-biliary fistula, Biloma, Embolization

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## Background

Pseudoaneurysm and arterio-biliary fistulas are rare complications of liver trauma. In a liver trauma patient, they evolve immediately secondary to direct vascular injury and usually present with either drop in hemoglobin level or hemobilia due to bleeding into the peritoneal space or biliary system. In the present case, there were no clinical features of hemobilia or upper gastrointestinal bleeding. Moreover, the hepatic artery pseudoaneurysm and arterio-biliary fistula evolved after a latent period of a few weeks, secondary to the toxic effects of the bile on the vessel walls due to biloma formation or biliary leakage, which is exceptionally uncommon. Multiple case reports and case series mentioning the pseudoaneurysm and arterio-biliary fistula have been described previously in the setting of liver trauma; however, delayed appearance of hepatic artery pseudoaneurysm and arterio-biliary fistula due to the toxic effects of the bile on the vascular walls secondary to biloma or bile leakage in a trauma patient is rarely described. This article will also give insight into the need for close monitoring and follow-up of these trauma patients for early detection and treatment of pseudoaneurysm and arterio-biliary fistula, especially

when there are clinical or imaging features of bile leakage.

## Case presentation

A 19-year-old male patient with a history of road traffic accidents presented to our emergency department. His vitals were stable at the time of presentation, with tenderness in the right upper quadrant. e-FAST (Extended Focused Assessment with Sonography in Trauma) was positive. The patient was further evaluated with contrast-enhanced computed tomography (CECT), which demonstrated a large contusion of the right lobe of the liver (Grade IV American Association for the Surgery of Trauma liver injury scale) involving the Couinaud segment VI, VII, and VIII with no obvious evidence of any pseudoaneurysm or active contrast extravasation. A gross hemoperitoneum was noted (Fig. 1). The rest of the abdominal organs were normal. As the patient's vitals were stable, the patient was kept for non-operative conservative management. On the 7th day of conservative management, his hemoglobin dropped by 2 units, and the patient was immediately shifted for digital subtraction angiography (DSA). Selective and superselective angiography of the right hepatic artery and its branches,



**Fig. 1** CECT axial (a) and coronal (b) venous phase images on day 1 of admission show a large contusion (thick white arrows) of the right lobe of the liver with gross hemoperitoneum (asterisk). CECT coronal arterial phase image (c) showed no pseudoaneurysm or active contrast extravasation from the right hepatic artery (RHA). Selective and superselective DSA angiography of the RHA and its branches (d, e) on the 7th day of conservative management when his hemoglobin dropped by 2 units, revealed no pseudoaneurysm or active contrast extravasation, so temporary embolization (f) of the RHA was done using Gelfoam

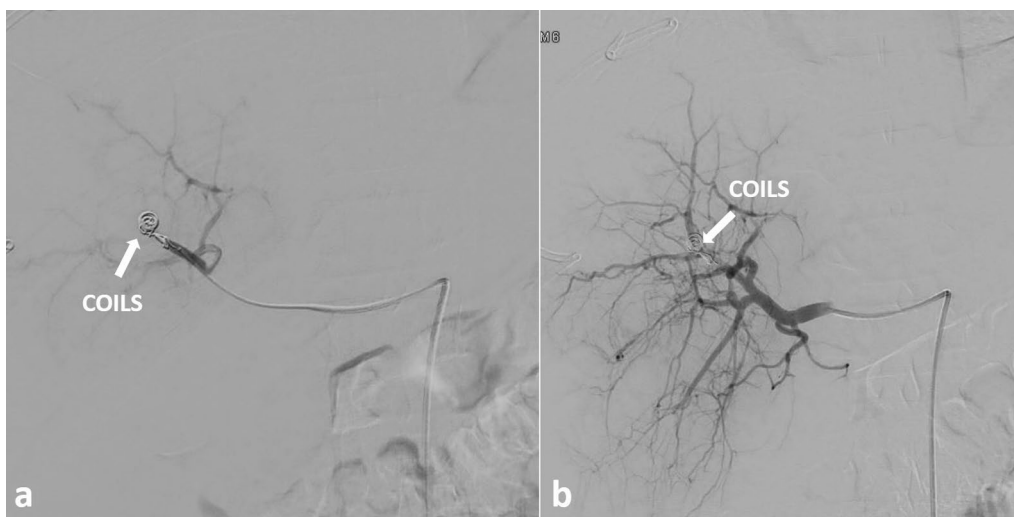
respectively, was done. However, no pseudoaneurysm or active contrast extravasation was noted, so temporary embolization of the right hepatic artery was done using Gelfoam (Fig. 1). The next day, the hemoperitoneum was also drained by putting a pigtail drainage catheter in the pelvis. Close monitoring was done, and no further drop in the hemoglobin was noted. The pigtail drainage catheter was removed after 2 days (the 10th day of admission) when the entire hemoperitoneum was drained, confirmed on ultrasound. On the 15th day of the admission, he gave a history of mild persisting right upper quadrant discomfort. On examination, he had a fever, and swelling was noted in the right upper quadrant region, so an ultrasound was done, which demonstrated a large well-defined anechoic collection in the perihepatic space overlying the right lobe of the liver. On ultrasound, there was no dilatation of intrahepatic biliary channels or intraluminal echogenic material within the biliary system. The collection was drained with a pigtail drainage catheter, which was bilious in nature and consistent with biloma formation/biliary leak. The pigtail drainage of the collection was both diagnostic and therapeutic in nature. Approximately 1 Liter of bilious fluid was drained. The catheter continued to drain 150–250 ml of bile per day. On the 23rd day of the admission, his hemoglobin again dropped with evidence of blood-mixed bile in the drainage bag. There was no history of hematemesis or malena. So, an emergent CECT was done, which showed a small pseudoaneurysm arising from the anterior segmental branch of the right hepatic artery (Fig. 2). No active contrast extravasation or features of the arterio-biliary fistula was noted on CECT. The patient was again shifted to the DSA suite for embolization of the pseudoaneurysm after interventional radiology consultation. Using a 5 Fr RC-1

catheter (Rosch Celiac, Cook, USA), angiography of the proper hepatic artery was taken, demonstrating a small pseudoaneurysm arising from a segmental branch of the right hepatic artery (Fig. 2). The culprit branch was super-selectively catheterized using the microcatheter (Progreat, Terumo, Japan). A repeated contrast angiography demonstrated the hepatic artery pseudoaneurysm with active contrast extravasation and opacification of the biliary system in the arterial phase consistent with the arterio-biliary fistula as additional findings (Fig. 2). First, the aneurysmal sac was filled with 2 embolization coils (Nester, Cook, USA), followed by the proximal closure of the culprit arterial branch with 1 coil (Nester, Cook, USA). Post-procedure, there was no evidence of pseudoaneurysm or active bleeding with the closure of the arterio-biliary fistula (Fig. 3). The drain from the pigtail catheter was reduced to 50 ml daily and became nil after 8 days of coil embolization. The pigtail draining catheter was removed, and the patient was discharged in stable condition after 10 days of coil embolization (33<sup>rd</sup> day of admission). At 1 monthly follow-up, the patient was asymptomatic and doing well with no intra-abdominal collection, pseudoaneurysm, or dilated biliary channels on ultrasound.

Pseudoaneurysms and arterio-biliary fistulas are infrequent complications of surgery or iatrogenic trauma (cholecystectomy, percutaneous biliary drainage, liver biopsy, endoscopic procedures), accidental trauma, tumors, vascular malformations, infection, and inflammation of the biliary tract [1, 2]. They usually present with hemobilia; however, our patient had no clinical features of hemobilia; instead, we had biloma formation and a drop in hemoglobin level. Pseudoaneurysms and arterio-biliary fistulas are formed due to direct injury to



**Fig. 2** CECT coronal arterial phase image (a) on day 23 of admission showed a small pseudoaneurysm (white arrow) arising from the segmental branch of the right hepatic artery (RHA). A draining catheter is seen in the right perihepatic space (asterisk) for biloma drainage. No active contrast extravasation or features of the arterio-biliary fistula was noted on CECT. DSA angiography of the RHA (b) revealed a small pseudoaneurysm (white arrow) arising from a segmental branch of the RHA. The culprit segmental branch was super-selectively catheterized using the microcatheter, and a repeated contrast angiography (c) demonstrated the hepatic artery pseudoaneurysm (white arrow) with active contrast extravasation (thick black arrow) and opacification of the biliary system (thin black arrows) in the arterial phase consistent with the arterio-biliary fistula



**Fig. 3** Post-coil embolization (a, b), there was no evidence of pseudoaneurysm or active bleeding with the closure of the arterio-biliary fistula

the vessel or toxic effects of the bile on the adjacent vessel walls secondary to bile leakage [3, 4]. In our case, the delayed appearance of the hepatic artery pseudoaneurysm and the arterio-biliary fistula can be explained by the toxic effects of the bile on the adjacent vessel walls secondary to bile leakage and biloma formation.

Hemobilia can present after several months of trauma with symptoms such as right hypochondrium pain, jaundice, and upper gastrointestinal hemorrhage depending upon the amount and velocity of the bleeding within the biliary system [5].

For patients with upper gastrointestinal hemorrhage, upper gastrointestinal endoscopy is the first modality to be used because it can detect blood clots in the ampulla of Vater and rule out other causes of bleeding. Ultrasound is a helpful method to detect hemobilia by detecting blood clots or echogenic intraluminal material within the biliary channels or gallbladder. CECT can reveal intrahepatic pseudoaneurysms, dilated bile ducts, and hyperdense intraluminal material within the biliary system consistent with arterio-biliary fistula [6]. Additionally, CECT is also helpful in planning angiographic interventions by assessing the vascular anatomy. The gold standard to detect pseudoaneurysm and arterio-biliary fistula is angiography. It offers diagnostic and therapeutic options to manage the bleed through coil embolization or stent deployment [7]. Moreover, it helps preserve maximum liver parenchyma function through selective embolization of the appropriate vascular branches.

Irrespective of the cause, endovascular hepatic artery embolization is the treatment of choice for managing intrahepatic pseudoaneurysms and arterio-biliary fistulas due to its 80–100% success rate with low morbidity and

mortality rates [2–4, 8, 9]. But there is a risk of developing an intrahepatic abscess or biliary strictures following embolization [8]. In the case presented here, the patient underwent selective transcatheter embolization of the hepatic artery pseudoaneurysm and the culprit branch of the right hepatic artery. Surgical hepatic artery ligation and liver resection are reserved for those cases where endovascular treatment is unsuccessful or not feasible [10].

## Conclusions

In liver trauma patients, the latency period for developing a pseudoaneurysm and an arterio-biliary fistula can be from a few days to a few months due to direct vascular injury or toxic effects of the bile on the adjacent vessel walls secondary to bile leakage or biloma formation. Development of hepatic artery pseudoaneurysm and an arterio-biliary fistula after a latent period of a few weeks, secondary to the toxic effects of the bile on the vessel walls due to biloma formation or biliary leakage, is exceptionally uncommon. So, these liver trauma patients require close monitoring and follow-up for early detection and treatment of pseudoaneurysm and arterio-biliary fistula, especially when there are clinical or imaging features of bile leakage or biloma formation. Endovascular treatment of hepatic artery pseudoaneurysm and arterio-biliary fistulas is a safe and effective method.

## Abbreviations

e-FAST	Extended Focused Assessment with Sonography in Trauma
CECT	Contrast-enhanced computed tomography
DSA	Digital Subtraction Angiography
RHA	Right hepatic artery

**Acknowledgements**

Not applicable.

**Author contributions**

Conceptualization, original draft preparation: BS, PKS; Review and editing: MKN, PK, PKS, and KM. All authors have read and approved the manuscript.

**Funding**

Not applicable.

**Availability of data and materials**

Not applicable.

**Declarations****Ethics approval and consent to participate**

Not applicable.

**Consent for publication**

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the consent form is available for review by the Editor of this journal.

**Competing interests**

The authors declare that they have no competing interests.

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Received: 15 May 2023 Accepted: 18 November 2023

Published online: 27 November 2023

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