

CASE REPORT

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A rare presentation of haemobilia with acute pancreatitis: a case report

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Abstract

Background Haemobilia, caused by hepatic artery aneurysms leading to acute pancreatitis is a rare presentation. Pancreatitis is hypothesized to result from a similar mechanism to gallstone obstruction at the ampulla of Vater. Herein, we report a large common hepatic artery (CHA) and right hepatic artery (RHA) aneurysm with a common bile duct (CBD) fistula causing acute pancreatitis and upper gastrointestinal bleeding.

Case presentation A 61-year-old male, with nicotine use disorder and hypertension, presented to the emergency department for the first time with a sudden onset of severe epigastric pain radiating to the back, relieved by bending forward, and two episodes of haematemesis and melena. His laboratory findings showed anaemia, elevated amylase and bilirubin with predominant direct component. Imaging revealed a partially thrombosed fusiform CHA aneurysm extending to the RHA and porta hepatis with a fistula into the CBD. Following stabilization, operative management was carried out, which included CBD exploration with clot evacuation, fistula oversewing, cholecystectomy, Roux-en-Y hepaticojejunostomy, and ligation of the common hepatic artery proximal to the aneurysm after ensuring the presence of a replaced left hepatic artery and collateral supply to the liver. One-year post-operative follow-up angiogram showed complete thrombosis of the aneurysm with preserved hepatic perfusion.

Conclusions This case highlights haemobilia as a rare cause of acute pancreatitis, emphasizing the need for high clinical suspicion and timely surgical intervention.

Keywords Acute pancreatitis, Haemobilia, Hepatic artery aneurysm, Arterio-biliary fistula

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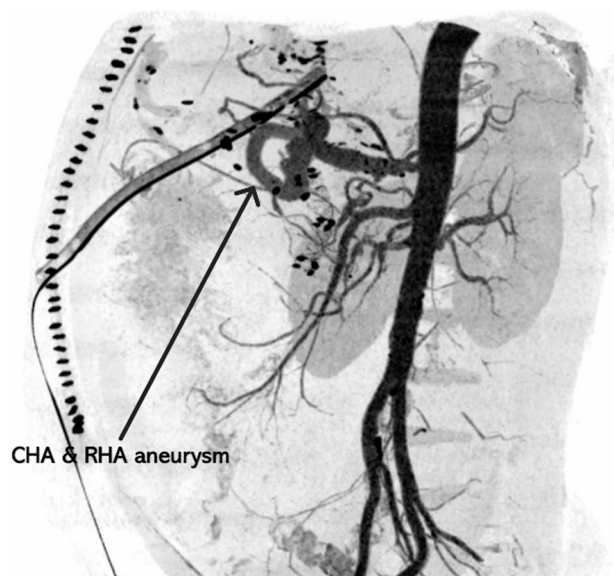


Fig. 1 CT angiogram demonstrating common hepatic artery aneurysm extending to right hepatic artery and porta-hepatis before the ligation of the aneurysm (CHA: common hepatic artery, RHA: Right hepatic artery)

Background

Haemobilia is abnormal bleeding into the biliary duct system by a fistula between the vasculature and duct system, the majority resulting from hepatobiliary procedures or trauma [1]. Haemobilia by hepatic artery aneurysms has a reported incidence of 2 in 100,000 and is observed during the fifth to sixth decade of life [2]. Herein, we report a rare presentation of acute pancreatitis caused by haemobilia secondary to a common hepatic artery (CHA) aneurysm with a common bile duct (CBD) fistula.

Case presentation

A 61-year-old male with nicotine use disorder, diagnosed with hypertension with good control, presented to the emergency department with an acute epigastric pain radiating to the back with two episodes of haematemesis and melena for the first time. There was no history of epigastric pain, trauma, abdominal surgeries or procedures. He denied of alcohol use. He was pale with no jaundice or fever. On admission he had a Glasgow coma scale of 15/15, pulse rate of 110/min, blood pressure of 103/68mmHg and was clinically pale. His abdomen was tender in the epigastric region, with no distention, palpable masses or evidence of free fluid. The diagnosis of acute pancreatitis was based on the modified Atlanta classification with two out of the three criteria fulfilled. On admission his haemoglobin was 8.9 g/dl, white cell count $15.2 \times 10^9/\text{ul}$, C-reactive protein 139.7 mg/L, amylase 800IU/L, total bilirubin 2.1 mg/dl, direct component 1.5 mg/dl, Alkaline phosphatase 284IU/L, Gamma-glutamyl transferase 739IU/L and elevated transaminases: (Alanine Aminotransferase(ALT) 231U/L and Aspartate

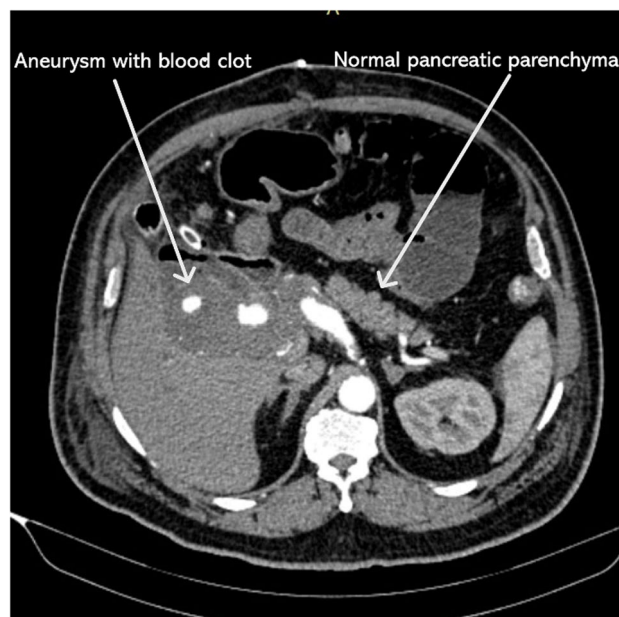


Fig. 2 Contrast-enhanced CT abdomen demonstrating a large common hepatic artery aneurysm causing pressure effects at the porta-hepatis. The gallbladder is filled with enhancing material suggestive of blood. The pancreas demonstrates no evidence of pancreatitis

Aminotransferase (AST) 53U/L). His prothrombin time (PT) was 15.2 s and INR 1.09. After initial resuscitation with normal saline 2 units and 2 units of blood transfusions, the patient was haemodynamically stabilized. Ultrasonography revealed a CHA aneurysm. This was confirmed by the Computed Tomography scan (CT) of the abdomen with angiogram, which revealed a partially thrombosed fusiform aneurysm of the CHA extending into the RHA and up to the porta hepatis (Fig. 1). The mass effect of aneurysm caused mild intrahepatic duct dilatation without CBD dilatation, and the cystic duct appeared incorporated into the aneurysm and the gallbladder was filled with enhancing material suggestive of blood (Fig. 2). Magnetic Resonance Cholangiopancreatogram (MRCP) with Magnetic Resonance Imaging (MRI) confirmed the CT findings. A cystic duct to the CHA aneurysm was suspected following initial imaging. After a multidisciplinary team discussion with a suspected CHA-cystic duct fistula and considering the larger size of the aneurysm and patient instability, a decision was made to do an open cholecystectomy with CBD stenting. There was no active bleeding during the first surgery. The gallbladder was found to be full of blood clots, thus an assumption was made that the fistula was from the CHA to the cystic duct. As the CBD also had blood clots, on table Endoscopic Retrograde Cholangio Pancreatogram (ERCP) and CBD clearance done followed by a stent placement inside the CBD (Fig. 3). Due to the presence of a vasculo-biliary fistula, a covered stent was placed as a temporizing method to cover the fistula site.

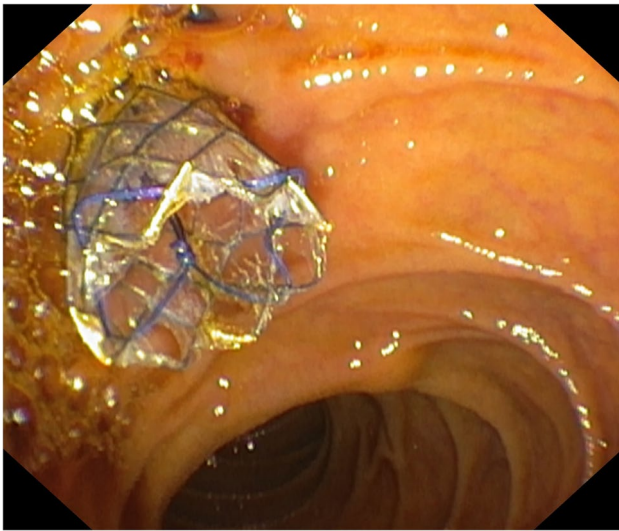


Fig. 3 Covered metal stent placed into the common bile duct after clot evacuation

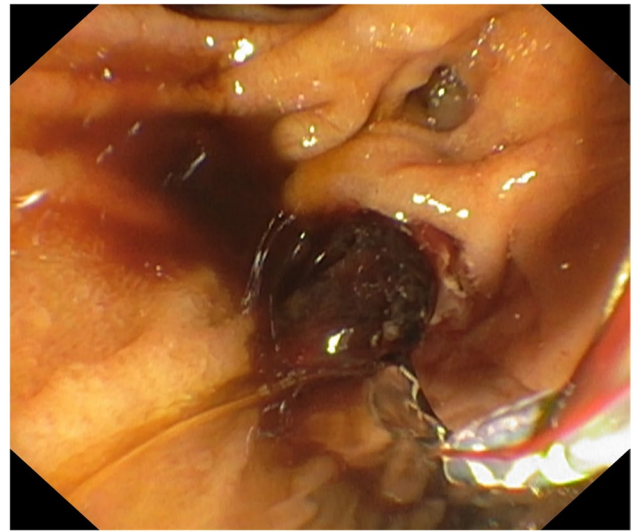


Fig. 5 Clot obstructing the stent placed into the common bile duct



Fig. 4 Post cholecystectomy sample demonstrating gallbladder filled with blood clots

Post-operative cholecystectomy sample demonstrating the gallbladder filled with blood clots is shown in Fig. 4. The patient improved initially with the surgery, two days after the procedure the patient became haemodynamically unstable with upper gastrointestinal endoscopy demonstrating clots obstructing the metal stent, which suggested persistent haemobilia (Fig. 5).

Re-exploration laparotomy was performed following massive transfusion. CHA to CBD fistula identified at the posterior wall of the CBD and oversewn with 2/0 polypropylene. The CBD was excised and hepaticojejunostomy was performed. An intraoperative decision was made to ligate the CHA at its origin after confirming the presence of a replaced left hepatic artery arising from the celiac axis. Ligation was carried out using 0 polyglactin sparing the left hepatic and splenic arteries, to prevent recurrence of the aneurysm. On-table hepatic duplex showed satisfactory flow in the left hemiliver with some arterial flow in the right liver and absence of the CHA aneurysm. His post-surgical follow up CT angiogram at one week (Fig. 6) revealed preserved arterial flow into the left lobe of the liver and collateral supply to the right lobe artery via segment IV branch. Smoking cessation was reinforced before discharging the patient to prevent future recurrences of aneurysm, development of cardiovascular events and chronic pancreatitis. One-year post-operative CT angiogram revealed a satisfactory outcome with complete thrombosis of the aneurysmal dilatation of RHA without residual filling and preserved flow in the right lobe via the collateral segmental supply from segment IV branch without any evidence of liver infarction.

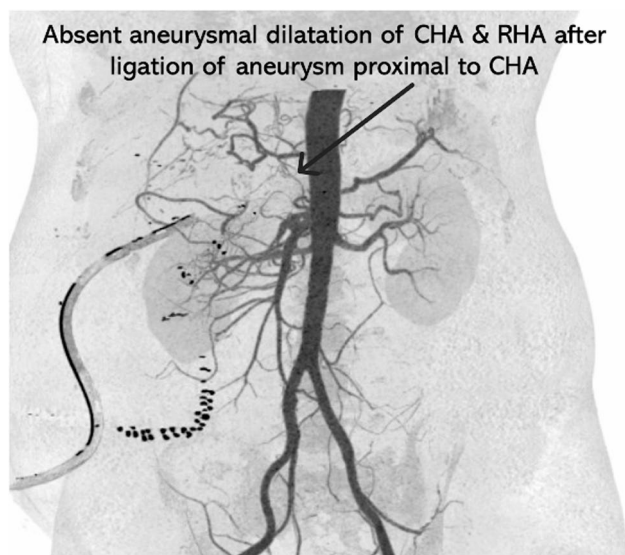


Fig. 6 One-week post-surgical CT angiogram demonstrating the satisfactory liver blood flow with no recurrence of the aneurysm after ligation of the aneurysm proximal to the common hepatic artery dilatation (CHA: common hepatic artery, RHA: Right hepatic artery)

Discussion

Sandblom in 1948 first introduced the term ‘haemobilia’ as bleeding into the biliary duct system after liver injury, a term currently in use for causes with blood in the biliary duct system [3]. Haemobilia may result from iatrogenic causes (60%), blunt or penetrating trauma, CBD stones, gallbladder carcinoma, CHA and liver abscesses [4]. Pancreatitis is rarely caused by an obstructing thrombus, which is hypothesized to result from a similar mechanism to gallstone obstruction at the ampulla of Vater by the pressure on the pancreatic duct. Pancreatitis being a common surgical presentation, and due to the rarity of having a vasculo-biliary fistula with haemobilia causing pancreatitis needs a high degree of suspicion.

The majority (80%) of the hepatic artery aneurysms are extrahepatic, with 63% involving the CHA, 28% RHA and 5% left hepatic artery [5]. Most of these aneurysms are asymptomatic prior to rupture [5]. Rupture may occur into the peritoneum or biliary tree in similar frequencies, with life-threatening bleeding [6]. Bleeding into the biliary tree results in haematemesis, melena and thrombosis, which may cause obstructive jaundice [1]. However, the classical triad of epigastric pain, obstructive jaundice and haemobilia is seen in only 30% [2]. Ruptures are common in those with non-atherogenic origin such as vasculitis, with aneurysm size over 2 cm and those with multiple hepatic artery aneurysms [7]. These aneurysms can be diagnosed using CT angiography and MRI demonstrating the size, shape and location [1]. The patient’s age, co-morbidities, size and location of the aneurysm largely determine the therapeutic strategies; thus, an individualized approach should be carried out. While percutaneous

embolization is useful in intrahepatic ruptured aneurysms, this technique has potential complications such as hepatic abscess, cholecystitis and long-term risk of re-canalization of the aneurysm [7, 8]. The first successful ligation of hepatic artery aneurysms was reported by Kehr in 1903 [9]. Ligation of CHA can be safely deployed by ensuring a patent gastroduodenal artery or pancreaticoduodenal artery with collateral blood flow to the liver [10]. In the absence of the collaterals, the potential risk of liver ischemia can be demonstrated by intra-operative occlusion of the CHA and observation for cyanosis; moreover, in the absence of collaterals revascularization should be attempted [10]. In this case as the patient was haemodynamically unstable, furthermore a clot was found inside the aneurysm and a covered vascular stent in the length of the aneurysm was unavailable. The decision was taken for open surgery after a multidisciplinary team involving a vascular surgeon and an intervention radiologist. Cholecystectomy was performed as the RHA was involved and there was a high possibility of gallbladder ischemia. Porto-biliary fistulas can be managed with stent placement of CBD [11]. However, in this instance as there was stent management failure with a large aneurysm causing CBD compression with recurrent haemobilia. Fistula was from the posterior wall of CBD to the anterior wall of the aneurysm. This was demonstrated after achieving proximal and distal circumferential control of the CBD. The fistulous tract was sutured with polypropylene. After these manipulations, in view of potential future risk of biliary strictures, a decision was taken to do a biliary bypass, which lead to CBD excision and Roux-en-Y hepaticojejunostomy.

Conclusions

Acute pancreatitis is a common surgical entity with a myriad of aetiologies. Haemobilia by a vasculo-biliary fistula can lead to pancreatitis through a similar mechanism to gallstone obstruction at the ampulla of Vater. Ligation proximal to a CHA aneurysm can be carried out as in this instance, when a replaced left hepatic supply is present, and after ensuring intra-operative adequate blood supply to the liver is maintained. High degree of clinical suspicion and urgent surgical interventions could reduce mortality in vasculo-biliary fistulas with a clinical presentation of acute pancreatitis.

Abbreviations

CHA	Common hepatic artery
RHA	Right hepatic artery
CBD	Common bile duct
CT	Computed Tomography
MRCP	Magnetic Resonance Cholangiopancreatogram
MRI	Magnetic Resonance Imaging

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Clinical trial number

Not applicable.

Authors' contributions

MN and NG performed the surgery. MN performed upper endoscopy. EG performed radiologic intervention. MN and JS prepared the manuscript. All authors (MN, GD, HM, YD, EG, NG, RP, JS) read and approved the final manuscript.

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Data availability

Not applicable.

Declarations**Ethics approval and consent to participate**

No ethical clearance required as it only involves a case report.

Consent for publication

Written informed consent was obtained from the patient for publication of clinical details, including radiological and procedure images. A copy of the consent can be provided upon request. Not applicable.

Competing interests

The authors declare no competing interests.

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