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Case Report

Endovascular treatment of a rare case of haemobilia caused by both pseudoaneurysm and a giant hepatic haemangioma [☆]

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ABSTRACT

Haemobilia is defined as bleeding from the biliary system due to abnormal communication between a blood vessel and the bile ducts. Melena or hematemesis, abdominal pain and jaundice represent the pathognomonic triad for haemobilia, but clinical presentation and aetiology of this entity are extremely variable. We report a case of a 50-year-old man with melena and anaemia and a clinical history of multivalvular endocarditis in which an extremely rare presence of 2 uncommon causes of haemobilia was found, such as a mycotic pseudoaneurysm and a giant hepatic haemangioma, both treated by transarterial embolization. In the management of haemobilia, TAE has been proven to be the treatment of choice because it combines a diagnostic angiography with therapeutic intervention in a minimally invasive, safe and effective way. Physician and radiologist should keep in mind also the uncommon aetiologies of haemobilia, knowing that the source of bleeding could be more than just one.

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Background

Haemobilia is defined as bleeding from the biliary system due to abnormal communication between a blood vessel and a bile duct.

The first to describe its symptoms was Glisson in 1654 [1]. In 1948, Sandblom [2] introduced the term "haemobilia" and described its pathognomonic triad: gastrointestinal bleeding (melena in 90% of patients, hematemesis in 60%), pain in the upper right quadrant (70%) and jaundice (60%). Moreover, in his review of 545 patients, Sandblom described the causes:

Abbreviations: TAE, transarterial embolization; CTA, computed tomography angiography; RHA, right hepatic artery; HAP, hepatic artery pseudoaneurysm; SMA, superior mesenteric artery; TACE, transarterial chemoembolization; TAL, lipiodolization.

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Fig. 1 – Axial CTA of the abdomen shows a pseudoaneurysm (arrows) in the hepatic segment V with typical homogeneous enhancement in the arterial phase (a) that persisted on the venous (b) and delayed phases (c) arising from an enlarged branch of the right hepatic artery.

bleeding from the liver parenchyma, from the bile ducts, from the gallbladder and the pancreatic gland. Trauma was the most common cause in 48%, followed by infection in 28%, gallstones in 10%, aneurysm in 7% and tumour in 5%.

Up to date, a frequent cause of haemobilia is iatrogenic, due to the increase in diagnostic and therapeutic hepatobiliary procedures. Open and laparoscopic biliary tract surgery, liver biopsy, transhepatic and endoscopic procedures as well as extracorporeal lithotripsy can cause mucosal lesions, biliovascular fistulas or pseudoaneurysm that can rupture into the hepatic bile ducts.

Hepatic abscess and parasitosis (eg, ascariasis, amoebiasis, echinococcosis, schistosomiasis) can cause parenchymal necrosis, cholangitis and pseudoaneurysm that can subsequently determine haemobilia. Stones in the extrahepatic biliary system can cause mechanical irritation of the mucosa and symptomatic haemobilia. Haemobilia can also occur in association with choledochal cysts, heterotopic gastric mucosa or haematological diseases such as sickle-cell anaemia.

Vessels inflammation and vascular malformation can cause haemobilia in case they develop communication with the biliary tree.

Srivastava et al. reported 6 cases of vasculitis associated with haemobilia caused by liver abscesses, polyarteritis nodosa and mycotic pseudoaneurysm [3].

Some kind of tumours, in particular malignant neoplasms of the liver, bile duct or pancreas, can bleed into the biliary system. However, haemobilia due to a benign neoplasm, such as liver haemangioma, is extremely rare [4–6].

In the management of haemobilia, transarterial embolization (TAE) has been proven to be a treatment of choice because it combines diagnostic angiography and therapeutic intervention in a minimally invasive, safe and effective way.

We present an extremely rare co-presence of 2 uncommon causes of haemobilia, a mycotic pseudoaneurysm and a hepatic haemangioma, both treated by TAE.

Case presentation

A 50-year-old man – with a clinical history of cerebral ischaemia, right hemiparesis, cognitive impairments and multivalvular endocarditis with "E. faecium" sepsis following an aortic dissection surgery – was admitted in the emergency unit for melena and anaemia (Hb 8 mg/dL).

A CT angiography (CTA) was performed, showing an irregular sac-shaped lesion with thin walls (34 mm in diameter) in the hepatic segment V (according to Couinaud's classification) with homogeneous blood pool enhancement in the arterial and delayed phases. The lesion was fed by a hypertrophic branch of the right hepatic artery (RHA). These findings strongly suggested a hepatic artery pseudoaneurysm (HAP) (Fig. 1). Furthermore, a large vascular mass (64 mm in diameter) was detected in the segments V and VIII with centripetal enhancement, also supplied by a branch of the RHA – findings consistent with cavernous haemangioma (Fig. 2). In non-enhanced phase, haemobilia was found along with dilatation of the right biliary system, the common bile duct and the gallbladder, without signs of active bleeding in the small bowel after contrast media administration (Fig. 3). The patient was hemodynamically stable. Thus, he was urgently transferred to the angiographic suite where – after preparation of a sterile field and subcutaneous injection of lidocaine hydrochloride as local anaesthesia – a right common femoral arterial access was gained through a 5Fr vascular sheath. A Sim1- 5Fr catheter was inserted on a hydrophilic wire (Terumo 180 cm) and advanced to the celiac artery and a selective arteriography was performed.

Super-selective arteriography of the RHA – gained with a coaxial 2.7 Fr microcatheter – showed the HAP.

The HAP was embolized using 0.018" detachable microcoils (Penumbra Inc.) with the "sac-packing" technique. During the procedure, an opacification of the biliary system was detected, suggestive for an arterio-biliary fistula. The front-door vessel was also embolized with microcoils (Penumbra Inc). The final angiographic control showed the occlusion both of the pseudo-aneurysmatic sac and the feeding vessel (Fig. 4).

No retrograde supplies or sign of extravasation were found from the angiographic study of the celiac artery and of the superior and inferior mesenteric arteries. No peri- or post-procedural complications occurred.

A month later, the patient was re-admitted to the emergency unit due to recurrence of anaemia and melena. Therefore, a new CTA was performed, showing haemobilia despite the complete embolization of the HAP. From these radiological findings and the persistent haemobilia, active bleeding caused by the cavernous haemangioma was suspected.

Thus, a new angiography of the celiac artery and RHA was performed. Using a microcatheter, the RHA and the arterial branch feeding the cavernous haemangioma was super-selectively catheterized and embolized using embosphere

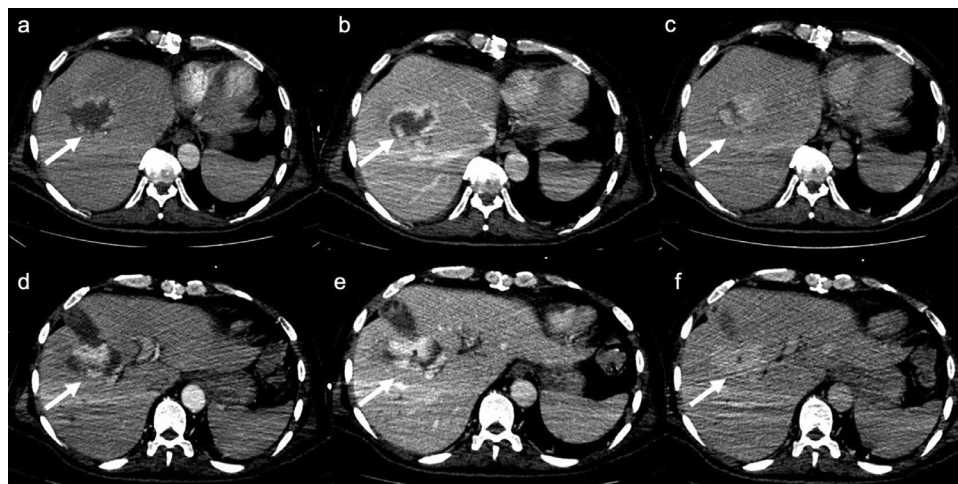


Fig. 2 – Axial CTA of the abdomen shows a large vascular mass (arrows) in the hepatic segments VIII (a-c) and V (d-f) with a progressive centripetal filling during the arterial (a,d), venous (b,e) and delayed phases (c,f) suggestive of cavernous haemangioma.



Fig. 3 – Axial non-enhanced CT shows haemobilia with blood dilatation and hyperdensity in the common bile duct (asterisk) and gallbladder lumen (arrow).

300-500 μm (Embogold - Merit Medical) and detachable microcoils (3 mm \times 3 mm). The control angiography showed a good morphological result (Fig. 5).

The arteriographic study of the superior mesenteric artery (SMA) was performed, showing no retrograde supplies or sign of extravasation. No peri- or post-procedural complications occurred.

After the treatment, haemobilia resolved and the haemoglobin value and the other vital's parameters improved. Therefore, the patient was discharged some days later.

Discussion

The clinical presentation of haemobilia depends on the rate of blood loss and it is extremely variable. Very fast arterial

bleeding can cause massive blood flow into the bile and thus into the duodenum, causing hematemesis or melena. Massive bleeding may also cause a sudden dilatation of the extrahepatic bile ducts leading to colic pain or even shock and death in the most severe scenarios.

When bleeding occurs more slowly, blood and bile do not mix easily. Thus, clots may form causing biliary obstruction with biliary colic and jaundice. Haemobilia from a venous source is rare [3,6].

There is a balance in the virtual space where bile and blood communicate between the feeding vessel pressure and the biliary duct pressure: the increase of the blood pressure wins against the biliary pressure, causing the dilation of these arterio-biliary fistulas and the transition of blood into the biliary system. Instead, when the blood pressure drops, the bleeding temporarily stops and the blood may coagulate causing clots that may obstruct the fistula. This process explains the intermittent clinical features of haemobilia. The intervals between episodes can last weeks, months or even longer. However, the disease rarely stops spontaneously [6].

Haemobilia is often diagnosed late and could be treated inadequately for a long time, thus the physicians should be aware of the various causes and presentations of haemobilia, including the rarest ones.

Foremost, haemobilia must be suspected after excluding more common bleeding sources in the gastrointestinal tract. Therefore, the first diagnostic procedure should be an esophagogastroduodenoscopy. Despite this, in the suspicion of haemobilia, the selective angiography of the hepatic artery remains the single most accurate test to reveal the bleeding source. Even if the passage of contrast media into the biliary tract is seen in only 25% of the tests performed, arteriography usually defines and localizes the arterial lesion, like arterio-biliary fistula, arterio-portal fistula, and pseudoaneurysm [3,6].

In the management of haemobilia, the patient's age and general condition must be considered, as well as the cause, site and severity of the bleeding.

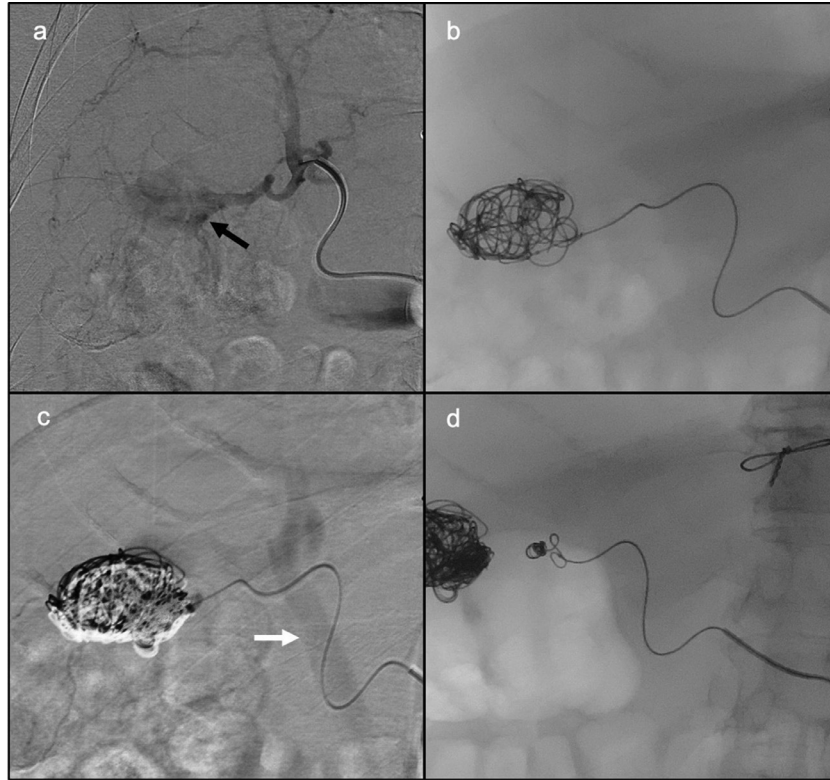


Fig. 4 – Super-selective arteriography by microcatheter of a branch of the RHA (a) showed the HAP (black arrow). Angiography images showing an embolization with micro-coils of the HAP arising from the RHA using a "sac-packing" technique (b) and occluding the front door (d). During the procedure, a transition of contrast medium into the biliary tree (white arrow) was found (c), suggestive for an arterio-biliary fistula.



Fig. 5 – Angiographic acquisition (a) and the super-selective angiography of the RHA by microcatheter (b) show a large cavernous haemangioma (arrow) fed by 2 branches of the RHA. After embolization of target vessels with embosphere 300-500 μm and micro-coils (c), the final angiographic control by the RHA (d) shows a good morphologic result with no more retrograde supplies or sign of extravasation.

To date, among the various therapeutic options, TAE is considered the treatment of choice compared to surgery because of its safety, mini-invasiveness and the possibility to combine diagnostic angiography with interventional treatment [3,6].

Super-selective catheterization of the feeding vessel of the bleeding source, followed by embolization with absorbable particulate matter seem to be most effective. Moreover, the risk of liver necrosis is minimal with super-selective embolization. Central embolization of the main hepatic artery or one of its principal branches is indicated whenever a peripheral branch cannot be catheterized or if the patient's clinical conditions mandate prompt and urgent intervention.

In case of pseudoaneurysms, the coils should be delivered at a site just distal to the arterial injury and extend across the lesion so that the neck of the aneurysm is bridged [3].

To our knowledge, there are only 3 cases of haemobilia caused by a cavernous haemangioma reported in the literature [4–6].

Haemangiomas are composed of endothelial cells from the hepatic artery, suggesting a place for vessel occlusive therapies such as TAE. To achieve greater tumour size reduction through the prevention of blood vessel growth, some chemotherapeutic agents may be administered in combination with TAE (transarterial chemoembolization or TACE). Moreover, chemotherapeutic agents can also be administered without vessel occlusive agents, for example, in combination with ethiodized oil (lipiodolization or TAL) [7].

In a recent review, Furumaya et al. compared different methods and materials for embolization of hepatic haemangiomas in eighteen cohort studies, including 1284 patients, proving the efficacy and safety of TAE/TAL as primary treatment for symptomatic hepatic haemangioma, regardless of the materials used.

When there is an indication for the treatment of liver haemangiomas (eg, large tumour size and/or symptoms), TAE/TAL appears to be a safe and effective treatment alternative to resection to decrease tumour size and also to provide relief of symptoms [7].

In our case, TAE has been proven to be a mini-invasive, safe and effective treatment of haemobilia that avoided major surgery.

Physician and radiologist should keep in mind also the uncommon aetiologies of haemobilia, knowing that the source of bleeding could be more than just one.

Patient consent

Written informed consent was acquired before all diagnostic exams and interventional procedures.

The patient's consent was obtained for the publication of this case.

The images in this case report are anonymized and are not accompanied by text might identify the individual concerned.

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