CASE REPORT

Spontaneous right hepatic artery branch gallbladder fistula revealed by haemobilia and upper cataclysmic gastrointestinal bleeding

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SUMMARY

Spontaneous right hepatic artery branch gallbladder fistula is a rare condition. Our case reported a spontaneous fistula between the right branch of the hepatic artery and the gall bladder. It constitutes a rare cause of haemobilia. In fact, the most common aetiology of haemobilia is traumatic or iatrogenic secondary to hepatobiliary surgery or interventions. Diagnosis of vascular-biliary fistula is not easy. The gallbladder endoluminal clot can mimic a mass, as in our patient. Selective arterial angiography is helpful in identifying the source of gastrointestinal haemorrhage. It can demonstrate the presence of arteriobiliary fistula. The differential diagnosis is arterial pseudoaneurysm in the vicinity of the vessel. Mini-invasive treatment of this fistula constitutes the best treatment. We here report a case of haemobilia with upper cataclysmic gastrointestinal bleeding revealing a spontaneous fistula between the right branch of the hepatic artery and the gall bladder.

BACKGROUND

Spontaneous right hepatic artery branch gallbladder fistula is a rare condition. We here report a case of haemobilia with upper cataclysmic gastrointestinal bleeding revealing a spontaneous fistula between the right branch of the hepatic artery and the gall bladder.

CASE PRESENTATION

A 35-year-old woman presented to the emergency department of our institution complaining of colicky pain in the right upper abdominal quadrant without fever. She had no history of traumatism, hepatobiliary surgery or intervention on the hepatobiliary tract. On admission, the patient was haemodynamically stable with a heart rate of 80 beats per minute, a blood pressure of 140/80 mm Hg and an oxygen saturation of 98%. Physical examination revealed jaundice and marked tenderness in the right upper abdominal quadrant. Laboratory results showed leucocytosis, elevation in total bilirubin, elevated gamma glutamyl transpeptidase and alkaline phosphatase-B, as well as a haemoglobin level of 13.4 mg/dL. Abdominal ultrasonography demonstrated a distended gall bladder with features compatible with althiasic cholecystitis. Abdominal CT revealed a gallbladder endoluminal mass that is spontaneously hyperdense (figure 1). It also showed

an important dilatation of the intrahepatic and extrahepatic bile ducts (figure 2).

While the exploration exams were being performed, the patient developed a cataclysmic upper gastrointestinal bleeding with haematemesis, rectal bleeding, haemodynamic instability and decreased haemoglobin level at 7 mg/dL. Emergency surgery was performed. An open surgical exploration demonstrated a haemobilia secondary to a fistula between the right branch of the hepatic artery and the gall bladder. This fistula caused the gall bladder to be filled with clots, explaining the endoluminal mass revealed by the CT. Closure of the fistula with preservation of the hepatic artery was performed, and involved Cholecystectomy and closure of the choledocotomy on a T-tube drainage were performed. The gall bladder was acalculous. The postoperative course was uneventful. Postoperative cholangiography showed fine biliary ducts without obstacle. The patient was discharged on the eighth postoperative day. Histopathological examination was consistent with acute and chronic cholecystitis involving all layers of the organ without signs of malignancy.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis is gallbladder tumour.

TREATMENT

Mini-invasive treatment of the fistula is recommended. It consists on an embolisation of the fistula with delayed laparoscopic cholecystectomy.

DISCUSSION

Our case reported a spontaneous fistula between the right branch of the hepatic artery and the gall bladder. It constitutes a rare cause of haemobilia.¹ In fact, the most common aetiology of haemobilia is traumatic or iatrogenic secondary to hepatobiliary surgery or interventions.²

Haemobilia is diagnosed via the pathognomonic Sandblom triad of haemobilia consists of upper abdominal pain, jaundice and gastrointestinal bleeding. . . Our patient initially presented with features of acute cholecystitis. She subsequently developed cataclysmic gastrointestinal haemorrhage.

Diagnosis of vascular-biliary fistula is not easy. The gallbladder endoluminal clot can mimic a mass, as in our patient. Selective transcatheter arterial

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Figure 1 Axial CT image showing an ill-defined mass formation within the gall bladder that is spontaneously hyperdense.

angiography is recommended to identify the exact location of the fistula. It also allows the embolisation of the feeder vessel with high accuracy. In our case angiography was not performed due to massive haemorrhage with haemodynamic repercussions. The differential diagnosis is arterial pseudoaneurysm in the vicinity of the vessel.

Mini-invasive treatment of this fistula constitutes the best treatment.³ It corresponds to an embolisation after angiographic localisation of the fistula with a delayed laparoscopic cholecystectomy.^{4,5} Surgery is used as salvage treatment in case of failed minimally invasive procedures, complications after these procedures, or their unavailability as in our case. Arterial ligation may lead to hazardous consequences on the liver: infarction, necrosis, abscess or duct strictures. Hence, repair of the vessel injury is recommended whenever it is possible.⁶ This repair varies from

Figure 2 Axial CT image illustrating dilated intrahepatic bile ducts.

closure, as in our patient, to end-to-end anastomosis or even graft interposition.⁷

In the literature, few cases of spontaneous fistula between cystic artery and gall bladder are reported.⁸ To our knowledge, no similar case of spontaneous right hepatic artery fistula into the gall bladder has been reported to date.

Priya *et al* reported a case of a spontaneous fistula between the cystic artery and the gall bladder. Angiography suggested a ruptured aneurysm in the vicinity of the right hepatic artery originating from the cystic artery with fistulous communication between the cystic artery and the gall bladder. Selective arterial embolisation was performed but failed because of coil migration. Surgical treatment was therefore needed, which consisted of retrograde cholecystectomy with right hepatic artery repair and retrieval of the microcoil.

Altmann *et al*⁹ reported a case of haemobilia revealing fistula caused by a corrosion of the cystic artery into the gall bladder.

Learning points

- ▶ Spontaneous fistula between the right branch of the hepatic artery and the gall bladder is a rare condition. Its diagnosis is challenging.
- ▶ Selective arterial angiography is helpful in identifying the source of gastrointestinal haemorrhage.
- ▶ Mini-invasive treatment of this fistula constitutes the best treatment.

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