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Case report

Hepatic artery pseudoaneurysm associated with cholecystitis that ruptured into the gallbladder

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Pseudoaneurysm of the hepatic artery due to cholecystitis may be very rare, and in our survey of the literature, the present case report is the first case of such a pseudoaneurysm. A 64-year-old woman presented with upper gastrointestinal bleeding and severe epigastric pain. Upper gastrointestinal tract endoscopy revealed blood coming out of the papilla of Vater. Color-Doppler ultrasound imaging showed a pulsatile wave pattern in an echogenic lesion inside the gallbladder. Contrast-enhanced computed tomography demonstrated a 3-cm pseudoaneurysm in the distended gallbladder. Angiography disclosed extravasation originating from the right hepatic artery. Emergency selective transcatheter arterial embolization was performed, with intravascular stainless steel microcoils, and complete occlusion of the pseudoaneurysm was achieved. The patient underwent cholecystectomy with resection of the extrahepatic bile duct and biliary reconstruction in a Roux-en-Y fashion. Macroscopically, the resected gallbladder contained clotted blood and multiple cholesterol stones. Microscopically, the mucosa of the gallbladder showed extensive necrosis and many inflammatory cells. The final diagnosis was pseudoaneurysm of the hepatic artery associated with calculous gangrenous cholecystitis. Although the mechanism of the pseudoaneurysm remains speculative, severe inflammatory reaction in the gallbladder may have infiltrated the liver parenchyma and may have eroded the wall of the hepatic artery, thus forming a pseudoaneurysm. Hemobilia is one of the important differential diagnoses when unexplained gastrointestinal bleeding is observed, especially in patients with hepatobiliary diseases.

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Introduction

Cholecystitis rarely causes pseudoaneurysm of the cystic artery or hemobilia. Only nine cases have been reported in the English-language literature.[1–9] However, pseudoaneurysm of the hepatic artery associated with cholecystitis is very rare, and to the best our knowledge, no published cases have been reported previously. In this report, we describe a rare case of hepatic artery pseudoaneurysm associated with cholecystitis that ruptured into the gallbladder. The patient was successfully managed by emergency selective transcatheter arterial embolization (TAE), with intravascular stainless steel microcoils, and subsequent surgery.

Case report

A 64-year-old woman presented with upper gastrointestinal bleeding and severe epigastric pain. The patient had had dull abdominal pain that had lasted for 2 weeks, and she had had a few episodes of hematemesis and melena. She had no prior history of hepatobiliary disease, abdominal trauma, or abdominal surgery. At the time of arrival, the patient was in shock. The laboratory data showed a decreased serum level of hemoglobin (6.1 g/dl), and elevated white blood cell count (16000/mm^3) and total bilirubin (3.9 mg/dl), with a direct fraction of 2.0 mg/dl. The patient received rapid fluid resuscitation and was stabilized. Emergency upper gastrointestinal tract endoscopy revealed blood coming out of the papilla of Vater. Ultrasound (US) showed multiple stones and an echogenic structure within the thick-

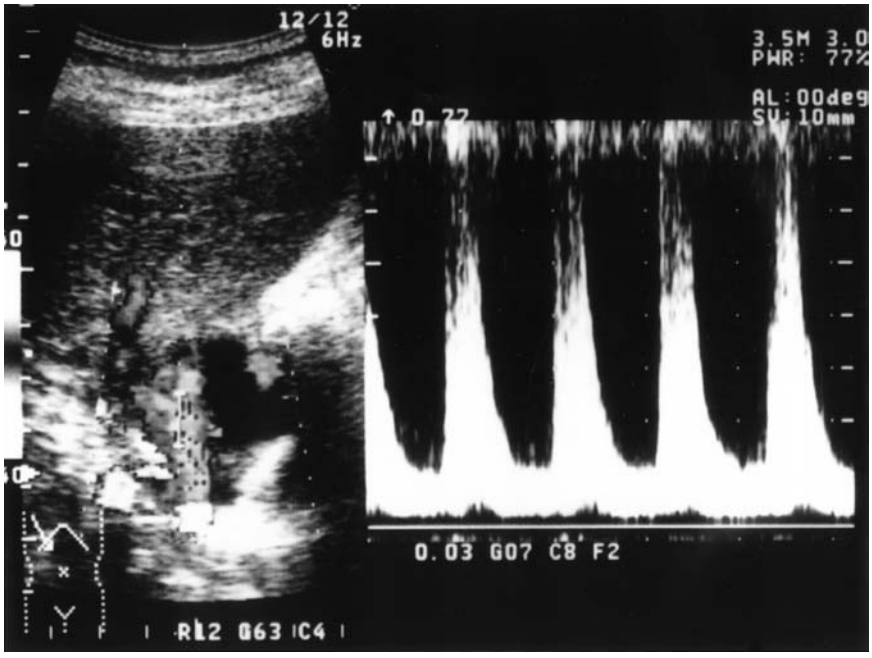

Fig. 1. Color-Doppler ultrasound (US) imaging showed a pulsatile wave pattern in an echogenic lesion inside the gallbladder

walled gallbladder. Biliary duct dilation was not observed. Color-Doppler imaging showed a pulsatile wave pattern in the echogenic lesion inside the gallbladder (Fig. 1). Contrast-enhanced computed tomography (CT) demonstrated a 3-cm pseudoaneurysm in the distended gallbladder (Fig. 2). Emergency angiography was planned, under the assumption of a pseudoaneurysm in the gallbladder. Selective angiography of the common hepatic artery disclosed extravasation originating from the right hepatic artery (Fig. 3). There were no abnormalities of the cystic arteries. Immediately after the diagnostic angiography, selective TAE was performed to control the hemorrhage. Intravascular stainless steel microcoils were placed in the proximal and distal portions of the origin of the aneurysm, using the Tracker Vascular Access System (Target Therapeutics, Fremont, CA, USA). Subsequent angiography showed complete occlusion of the pseudoaneurysm (Fig. 4) and the development of collateral pathways. Surgery was performed after a full evaluation, and when liver function had returned to normal. Surgical exploration demonstrated that the gallbladder was remarkably swollen and adhered strongly to the extrahepatic bile duct due to the inflammation of the gallbladder. There was no evidence of intraabdominal bleeding. The patient underwent cholecystectomy with resection of the extrahepatic bile duct and biliary reconstruction in a Roux-en-Y fashion. Vascular reconstruction of the right hepatic artery was not performed, because complete obstruction of the pseudoaneurysm had been achieved by TAE. Macroscopically, the resected gallbladder contained clotted blood and multiple cholesterol stones. Microscopically, the mucosa of the gallbladder showed extensive necrosis and many inflammatory cells. The clotted blood was surrounded by fibrous tissue with fibrinous exudates, and the arterial wall was not demonstrated within it (Fig. 5). The final diagnosis was pseudoaneurysm of the hepatic artery associated with calculous gangrenous cholecystitis. The patient recovered uneventfully and has been well, without recurrent bleeding, for 5 years.

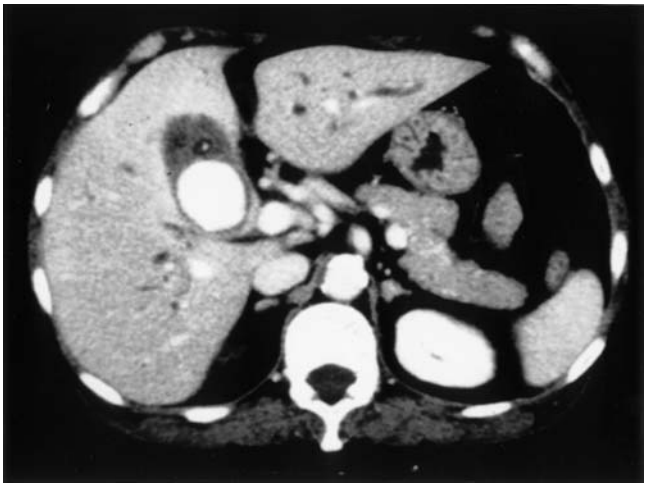

Fig. 2. Contrast-enhanced computed tomography (CT), demonstrating a 3-cm pseudoaneurysm in the distended gallbladder

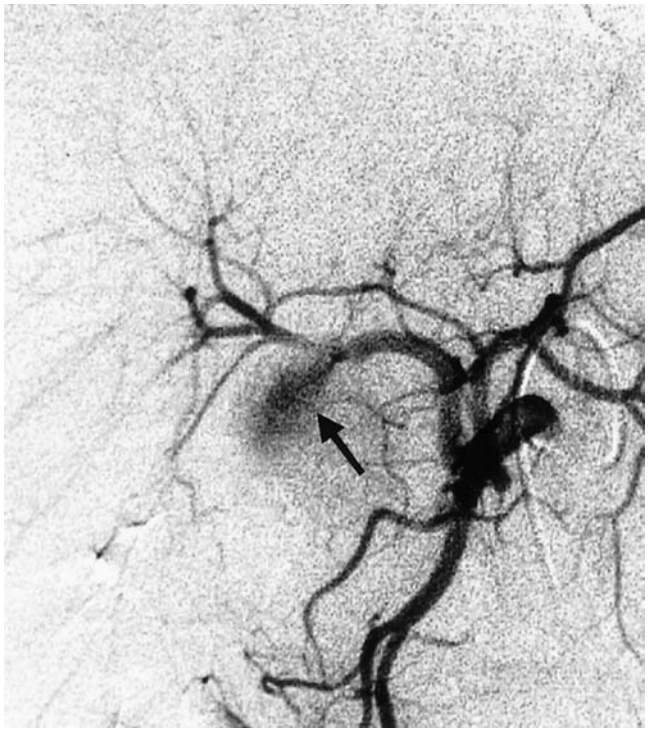

Fig. 3. Selective angiography of the common hepatic artery, disclosing extravasation (*arrow*) originating from the right hepatic artery

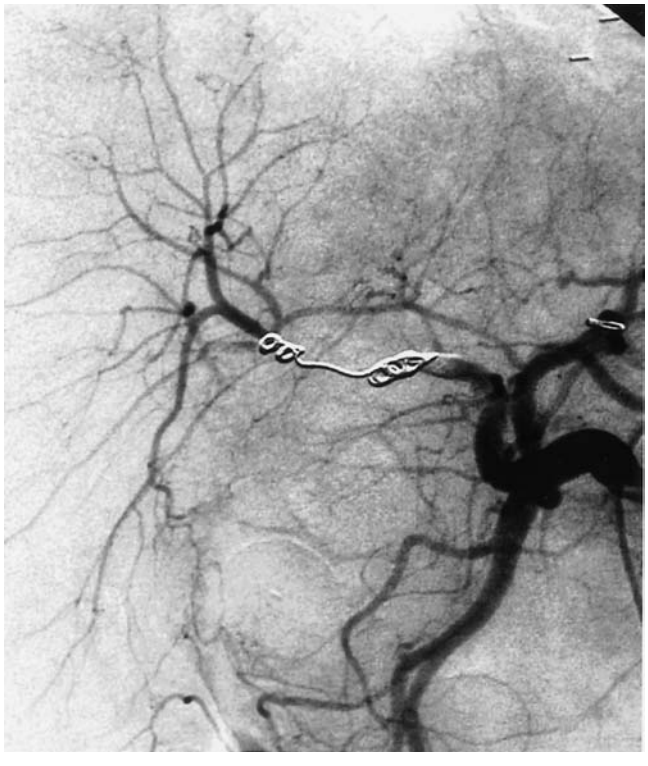

Fig. 4. Angiography after transcatheter arterial embolization (TAE) with intravascular stainless steel microcoils, showing complete occlusion of the pseudoaneurysm

Fig. 5. Clotted blood in the gallbladder. It is surrounded by fibrous tissue with fibrinous exudates, and the arterial wall is not demonstrated within it. H&E

Discussion

Pseudoaneurysm of the cystic artery is occasionally seen in patients with cholecystitis, and successful management by surgery and TAE with microcoils has been reported.[1–9] However, pseudoaneurysm of the hepatic artery that ruptured into the gallbladder may be very rare, and in our survey of the literature, the present case report is the first case. The detailed mechanism of the pseudoaneurysm remains speculative. The patient had cholelithiasis and gangrenous cholecystitis. This severe inflammatory reaction in the gallbladder may have infiltrated the liver parenchyma and may have eroded the wall of the hepatic artery, thus forming a pseudoaneurysm. The pseudoaneurysm ruptured into the gallbladder and caused hemobilia.

Sandblom[10] was the first to apply the term "hemobilia" to the syndrome of gastrointestinal bleeding due to hemorrhage into the biliary tract, and the syndrome consists of the classic triad of abdominal pain, jaundice, and upper gastrointestinal bleeding. Our patient had the triad, and upper gastrointestinal tract endoscopy demonstrated blood coming out of the papilla of Vater. However, many cases of hemobilia are diagnosed as unexplained gastrointestinal bleeding, and treatment tends to be delayed. Therefore, it should be emphasized that hemobilia is one of the important differential diagnoses when gastrointestinal bleeding is observed, especially in patients with hepatobiliary diseases.

Color-Doppler US has been reported to be very useful in detecting cystic artery pseudoaneurysms.[1,2] In the

present patient, US showed echogenic material within the gallbladder, and color-Doppler imaging demonstrated a persistent pulsatile flow within the echogenic lesion. These findings strongly suggested that the echogenic material could be a pseudoaneurysm in the gallbladder. CT clearly revealed a pseudoaneurysm in the gallbladder, and angiography showed extravasation originating from the right hepatic artery. Angiography can directly visualize a pseudoaneurysm and extravasation, and it offers significant information for diagnosis and treatment. Therefore, in patients with suspected pseudoaneurysm, we consider that angiography is mandatory prior to treatment.

Patients with ruptured pseudoaneurysm often develop shock, as occurred in the present patient. Because it is less invasive than other procedures, TAE has been reported to be effective in hemostasis in such patients.[11,12] There are several materials currently available for TAE. Gelfoam usually resolves within several weeks, whereas stainless steel microcoils produce permanent vascular occlusion;[11,12] thus, we used stainless steel microcoils. Moreover, the parental arterial wall of the pseudoaneurysm is generally fragile, and embolization of the aneurysm itself is associated with a danger of recurrent bleeding. Therefore, we performed embolization of the segmental parental artery. Liver infarction is an important problem after TAE. However, the liver can tolerate considerable embolization because of multiple collateral pathways, and the high selectivity of embolization contributes to the prevention of liver infarction. After TAE, our patient underwent delayed surgery. Such two-stage management allows patients to recover from hemorrhagic shock and liver dysfunction, and we can then accurately evaluate functional liver reserve.

References

1. Nakajima N, Hoshino H, Hayashi E, Nagano K, Nishimura D, Katada N, et al. Pseudoaneurysm of the cystic artery associated with upper gastrointestinal bleeding. J Gastroenterol 1996;31: 750–4.
2. Kaman L, Kumar S, Behera A, Katariya RN. Pseudoaneurysm of the cystic artery: a rare cause of hemobilia. Am J Gastroenterol 1998;93:1535–7.
3. Rhee JW, Bonnheim DC, Upson J. Cystic artery pseudoaneurysm. NY State J Med 1987;87:47.
4. Wu TC, Liu TJ, Ho YJ. Pseudoaneurysm of the cystic artery with upper gastrointestinal hemorrhage. Acta Chir Scand 1988;154: 151–2.
5. Strickland SK, Khury MB, Kiproff PM, Raves JJ. Cystic artery pseudoaneurysm: a rare cause of hemobilia. Cardiovasc Intervent Radiol 1991;14:183–4.
6. Barba CA, Bret PM, Hinchey EJ. Pseudoaneurysm of the cystic artery: a rare cause of hemobilia. Can J Surg 1994;37:64–6.
7. Reddy SC. Pseudoaneurysm of cystic artery with upper gastrointestinal hemorrhage. South Med J 1983;76:85–6.
8. Clements WDB, Wilson RH, Crothers JG, McIlrath EM, Johnston GW. Pseudoaneurysm of the cystic artery following cholecystectomy. J R Cool Surg Edinb 1993;38:348–9.
9. Bergey E, Einstein DM, Herts BR. Cystic artery pseudoaneurysm as a complication of laparoscopic cholecystectomy. Abdom Imaging 1995;20:75–7.
10. Sandblom P. Hemorrhage into the biliary tract following traumatic hemobilia. Surgery 1948;24:571–86.
11. Salam TA, Lumsden AB, Martin LG, Smith RB III. Nonoperative management of visceral aneurysms and pseudoaneurysms. Am J Surg 1992;164:215–9.
12. Mandel SR, Jaques PF, Sanofsky S, Mauro MA. Nonoperative management of peripancreatic arterial aneurysms. Ann Surg 1987;205:126–8.