Case report

Pseudoaneurysm of the cystic artery associated with upper gastrointestinal bleeding

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Abstract: Pseudoaneurysm of the cystic artery is a cause of hemobilia, and is extremely rare, with only eight cases having been reported in the world literature. We report a case of pseudoaneurysm of the cystic artery in a 72-year-old Japanese man. The patient experienced epigastric pain and melena, and was found to have jaundice and liver dysfunction. Repeated gastroendoscopy did not reveal the cause of the alimentary tract bleeding; however, color-Doppler ultrasonography detected an aneurysm of the cystic artery in the gallbladder. Selective hepatic arteriography demonstrated that the posterior branch of the cystic artery was markedly dilated and that an aneurysm had formed in the midst of the artery. We diagnosed hemobilia due to the pseudoaneurysm of the cystic artery, and associated gastrointestinal bleeding. Cholecystectomy was performed immediately. Pathologically, the gallbladder showed acute calculous cholecystitis. This case emphasizes the importance of including hemobilia in the differential diagnosis whenever gastrointestinal bleeding is associated with signs of biliary disorder; color-Doppler imaging is a favorable modality for the diagnosis of pseudoaneurysm of the cystic artery.

Key words: pseudoaneurysm, cystic artery, color-Doppler, hepatic arteriography

Introduction

Hematemesis or melena is a frequent clinical symptom of upper gastrointestinal bleeding. Its severity ranges widely and patients may exhibit such serious conditions as hypovolemic shock. Since more than 90% of upper gastrointestinal bleeding results from diseases of the alimentary tract such as peptic ulcers and varices, most cases can be easily diagnosed by emergent endoscopy. In patients with bleeding from regions other than the alimentary tract, it is difficult to confirm the precise source of bleeding by endoscopy alone. Hemobilia, hemorrhage into the biliary tract, is a rare cause of upper gastrointestinal bleeding, and is secondary to several other causes, such as hepatic trauma (accidental or iatrogenic), aneurysm of the hepatic artery, tumors of the biliary tract, gallstone disease, and inflammation of the liver. Pseudoaneurysm of the cystic artery is one of the causes of hemobilia, and is extremely rare, with only eight cases having been reported in the world literature. The condition may be associated with inflammation of the gallbladder; however, its cause remains obscure. Here we present a rare case of hemorrhage from the gallbladder caused by pseudoaneurysm of the cystic artery.

Case report

A 72-year-old Japanese man experienced melena and consulted his local hospital on January 26, 1995. He had had intermittent epigastric pain for a few weeks, and the pain had worsened after meals. At the hospital, jaundice and liver dysfunction were observed and he was therefore referred for further evaluation and admitted to our hospital, on January 30. He had a history of acute appendicitis at age 20 and intestinal tuberculosis at 32, and had no history of abdominal trauma.

The patient’s height was 152 cm and weight was 42 kg. Body temperature on admission was 36.8°C and blood pressure was 138/60 mmHg. On physical examination the bulbar conjunctiva showed slight icterus and the palpebral conjunctiva showed anemia. Inspection of the abdomen was unremarkable, except for an operation scar in the lower part; there was no tenderness over.
Fig. 1. Ultrasonography of the gallbladder. Abdominal ultrasonography shows distended gallbladder with an echolucent cystic lesion attached to the thickened wall. Doppler imaging detected pulsatile waves in this cystic lesion.

Fig. 2. Enhanced computed tomographic (CT) scan of the gallbladder. CT scan demonstrates a low-density lesion in the gallbladder, which is strongly enhanced by infusion of contrast medium.

The patient had melena. Physical examination was otherwise unremarkable.

Hematological values were: white blood cell count 6500/mm$^3$, red blood cell 230 $\times$ 10$^6$/mm$^3$, hemoglobin 7.3 g/dl, and platelets 48.8 $\times$ 10$^5$/mm$^3$. Coagulation test results were within normal limits. Blood chemistry showed the following abnormal values: total bilirubin 2.5 mg/dl (normal 0.2–1 mg/dl), direct bilirubin 2.2 mg/dl, aspartate aminotransferase 105 IU (normal 8–38 IU), alanine aminotransferase 233 IU (normal 5–33 IU), alkaline phosphatase 1980 IU (normal 88–288 IU), leucin aminopeptidase 262 IU (normal 30–64 IU), and $\gamma$-glutamyl transpeptidase 116 IU (normal 3–40 IU).

Gastroendoscopy was performed immediately on admission, revealing only for a small amount of "coffee ground" material in the stomach. No blood was detected in the duodenum. The patient received a transfusion of two units of packed red blood cells, and his condition remained relatively stable during the next few days. Gastroscopy was repeated, but no abnormalities were detected in the upper gastrointestinal tract. Abdominal ultrasonography showed a distended gallbladder with a few gallstones and an echolucent cystic lesion, 2.2 $\times$ 3.0 cm in diameter, attached to the thickened wall. Pulsatile waves were detected in the cystic lesion by color-Doppler imaging (Fig. 1). The common bile duct was not extended and no stone was observed in it by ultrasound imaging. Computerized tomographic (CT) evaluation of the abdomen demonstrated a well-defined cystic lesion, approximately 3 cm in diameter, in the lumen of the gallbladder, which was strongly enhanced by the infusion of contrast medium (Fig. 2). From these findings, we suspected hemorrhage into the gallbladder due to an aneurysm of the cystic artery, which had caused melena. Side-viewing duodenoscopy was performed, and bleeding from the papilla of Vater was confirmed on February 8, 1995. Subsequently, hepatic arteriography was carried out, revealing marked dilatation of the posterior branch of the cystic artery forming an aneurysm in the midst of the artery. Pulsatile jet flow of the contrast medium was followed into the aneurysm of the cystic artery by digital subtraction arteriography (DSA) (Fig. 3a). Pooling of contrast medium was recognized in the venous phase of DSA (Fig. 3b). We diagnosed pseudoaneurysm of the cystic artery as a cause of gastrointestinal bleeding, and cholecystectomy was performed on February 9.

Pathology evaluation showed a grayish-red gallbladder that was distended and contained turbid fluid, a giant hematoma, old blood clots, and two gallstones.