

A Rare Case of Spontaneous Gastrobiliary Fistula Diagnosed by Oral Contrast-enhanced Ultrasound

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Abstract: We report the case of a 55-year-old man with a spontaneous fistula between the intrahepatic bile duct and stomach. Oral contrast-enhanced ultrasound (CEUS) was used in this patient and made the diagnosis of fistula as observed during open surgery. We found that oral CEUS should be considered as a safe, simple and effective method to diagnose and evaluate gastrobiliary fistulas.

Key words: Gastrobiliary fistula; Contrast-enhanced ultrasound; Oral administration

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Gastrobiliary fistulas are quite rare. There were only a few cases reported by different centers [1–4]. Spontaneous gastrobiliary fistulas are rarer. Due to unclear clinical signs, false or delay in diagnosis usually happens. We reported a rare case of spontaneous fistula between the intrahepatic bile duct and stomach, which was diagnosed by oral CEUS.

Case report

A 55-year-old man was admitted because of recurrent abdominal pain for 6 months. The patient stated that significant abdominal pain had appeared a couple of times 10 years ago. However, he held off going to see a doctor, hence the diagnosis was not clear back then. Although the abdominal pain was not severe, he intended to figure out the reason for it. Physical examination after hospitalization showed that the abdomen was flat and soft. Slight tenderness and rebound tenderness were not felt. Murphy's sign was negative. No mass was palpated in abdomen. Levels of helicobacter pylori antibody and pepsinogen increased, and pepsinogen ratio decreased. Other laboratory tests, including blood tests, tumor biomarkers, liver function tests, etc. were normal.

Abdominal magnetic resonance imaging (MRI) combined with magnetic resonance cholangiopancreatography (MRCP) revealed mild dilatation of left intrahepatic bile duct with a small cyst near the left hepatic duct and chronic cholecystitis with multiple gallbladder stones. The anatomical structures of extrahepatic bile duct were normal (Fig. 1). No fistula was found.



Figure 1 Magnetic resonance cholangiopancreatography. The anatomy structures of the gallbladder, cystic duct, common hepatic duct and common bile duct are normal.

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Gastroduodenoscopy showed the presence of duodenal bulbar ulcers and chronic superficial erosive gastritis. A fistulous opening (10 mm × 3 mm) was found on the lesser curvature of the stomach near the pylorus (Fig. 2). Green and yellow bile adhered to the mucosa around the fistulous opening in the stomach, where no ulcers or masses could be seen after the bile was rinsed out. However, the exact route of the fistula could not be confirmed.

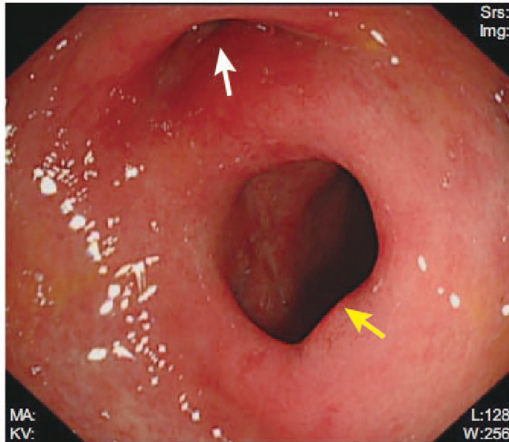


Figure 2 Gastroduodenoscopy. Image showing the opening (white arrow) of the fistula in the lesser curvature of the stomach and pylorus (yellow arrow).

Ultrasonography was performed by using Resona 7 ultrasound system (Mindray Medical Solutions, Shenzhen, China) equipped with an SC6-1 convex array probe (1-6 MHz). Dilatation and pneumobilia of the left intrahepatic bile duct were demonstrated (Fig. 3A), while the right intrahepatic bile duct and extrahepatic bile duct were not dilatated. The route of the common bile duct was normal. In addition, chronic cholecystitis with multiple gallbladder stones was also found. Considering the fistulous opening on the lesser curvature of the stomach found by gastroduodenoscopy, we attempted to use oral CEUS to visualize if there was fistula and identify its route. After preparation of a vial of SonoVue (Bracco, Italy) by mixing the contents with 5 mL of a 0.9% saline solution, 0.3 mL of contrast medium was diluted with 300 mL of water (SonoVue dilution concentration, 1:1000). Then, the patient was told to drink the dilute contrast agent. A low mechanical index (MI=0.067) was used for real-time CEUS. The contrast agent was observed to fill into the stomach and duodenal bulb. Meanwhile, the contrast agent flew through the stomach to the left intrahepatic bile duct, which demonstrated as a hyperechoic streak connected the lesser curvature of the stomach with the left intrahepatic bile duct (Fig. 3C). The patient was diagnosed with gastrobiliary fistula by oral CEUS finally.

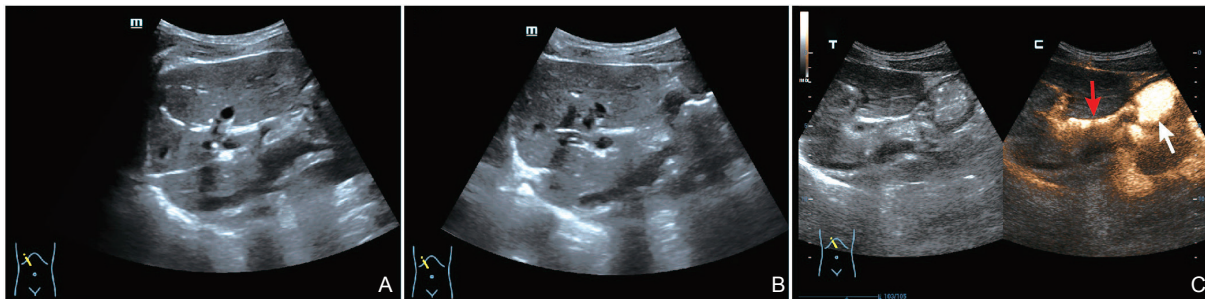


Figure 3 Abdominal ultrasonography. (A) Gray-scale ultrasonography showing pneumobilia and dilatation of the left intrahepatic bile duct; (B) Grayscale image before oral CEUS. The gastric organs and fistula could not be observed; (C) The left panel shows the fundamental mode, and the right panel shows the CEUS mode. The contrast agent was observed to fill in the stomach (white arrow). A hyperechoic fistula (red arrow) connected with the lesser curvature of the stomach and the left intrahepatic bile duct was detected.

The patient was transferred to the department of hepatobiliary surgery for further treatment. During the open operation, a channel-like structure connected the lesser curvature of the stomach with the left intrahepatic bile duct was found (Fig. 4A and 4B). No histopathological signs of inflammation with the fistula could be observed. Additionally, there were no tumors or gastric ulcers found. From the surgeon's point of view, the fistula seemed to be inherent and spontaneous. Considering the potential damage and possibility of malignant transformation of the left hepatic lobe, the left hepatic lobe was dissected during the surgery. The

opening of the fistula was observed clearly in the ex vivo left hepatic lobe (Fig. 4C). The gallbladder was also dissected for chronic cholecystitis with multiple gallbladder stones. The route of the fistula was confirmed between the lesser curvature of the stomach and the left intrahepatic bile duct. The fistula was cut and ligated at the distal part.

Discussion

Gastrobiliary fistula is not a common disease. It may develop as a result of liver ablation, tumor, and liver abscess [3-5]. Reported spontaneous gastrobiliary fistulas

are even more rare [1], the pathogenesis of which has not yet been clarified. Notably, the flow of bile into the

stomach through a fistula can cause gastric cancer [6,7]; therefore, it is important to make a clear diagnosis in time.

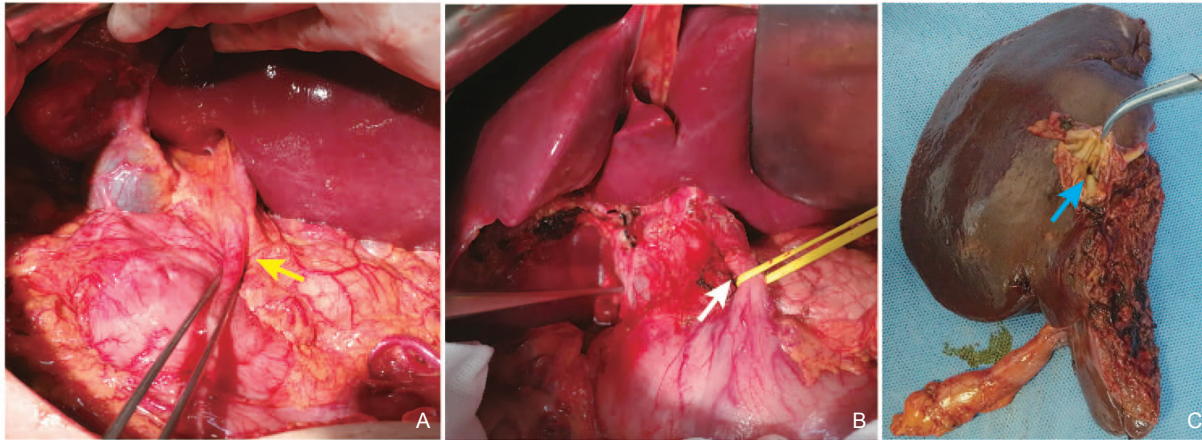


Figure 4 Findings during surgery. (A) The fistula (yellow arrow) was detected; (B) After the gallbladder was dissected and surrounding tissues were separated, the fistula between the lesser curvature of the stomach to the left intrahepatic bile duct (white arrow) was observed; (C) The opening of the fistula (blue arrow) was observed in the dissected left hepatic lobe.

Unclear clinical signs of gastrobiliary fistulas usually result in false or delay in diagnosis. In the cases of gastrobiliary fistulas reported previously, diagnoses were made by radionuclide imaging, contrast-enhanced computed tomography, intraoperative X-ray cholangiography or gastroduodenoscopy. The diagnosis remained uncertain until the operation. Moreover, there are disadvantages with these methods, such as radiation, invasiveness and high costs. In this case, MRI did not detect fistula, and gastroduodenoscopy could not confirm its route. Thus, non-invasive and accurate diagnostic methods are needed. CEUS has been considered as a reliable and safe tool. Its clinical value for the diagnosis, treatment and evaluation of liver and other organ diseases is approved by guidelines [8,9]. Most applications require intravenous injection of contrast agent. More remarkably, intracavitary contrast-enhanced ultrasound (IC-CEUS) is recommended for the delineation of cavities or ducts [10]. Injecting contrast agents through catheters into physiological or pathological cavities has been used in guiding percutaneous cholangiography and drainage [11,12], detecting peritoneal-pleural communication [13], guiding percutaneous nephrostomy [14], and managing postsurgical gastrointestinal fistulas [15]. Injecting contrast agent into one of the orifices of a fistula can directly improve visualization and is effective for the detection and classification of fistulas with CEUS [16,17]. However, there are only a few case reports about CEUS diagnosing fistulas, and all require catheter insertion, which is invasive and inconvenient. In this case, the upper gastrointestinal tract was taken as a tunnel to one of the orifices of the fistula. Thus, no catheter was needed. Oral CEUS played an important

role in diagnosing gastrobiliary fistula in this case. It could not only detect fistula but also defined its route noninvasively and expediently.

Oral contrast-enhanced gastric ultrasonography has been used in the detection and evaluation of gastric tumors and inflammatory bowel diseases [18,19]. Oral administration allows better visualization of the digestive lumen, especially in the upper gastrointestinal tract [20]. It improves diagnostic performance in discriminating gastric masses compared with conventional ultrasound [21,22]. Remarkably, in these studies, appropriate oral gastrointestinal iso-echoic ultrasound contrast agents such as herb mixtures (coix seed, yam, and pericarpium citri reticulatae) or polyethylene glycol were used. The echo of these oral gastrointestinal contrast agents was not clear enough to demonstrate fistulas. There were no reports about diagnosing gastric fistulas with these oral gastrointestinal contrast agents. On the other hand, even if SonoVue was diluted, it still worked better due to hyperechoic imaging under harmonic sonography. The excellent fluidity of SonoVue is more suitable for visualizing narrow ducts, such as fistulas. There were no adverse events.

So oral CEUS with diluted SonoVue appeared to be a non-radiative, noninvasive, convenient and effective method to diagnose gastrobiliary fistula.

Conclusion

We describe here, for the first time in the literature, a case of spontaneous fistula between the intrahepatic bile duct and stomach diagnosed by oral CEUS. Our case supports the notion that oral CEUS can be considered

as a safe, simple and effective method to diagnose and evaluate gastrobiliary fistulas.

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Conflict of Interest

All authors concur with the submission. There are no financial/commercial conflicts of interest related to this work.

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