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Intraoperative air leak test was useful for the detection of a small biliary fistula: A rare case of non-parasitic hepatic cysts with biliary communication

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ABSTRACT

INTRODUCTION: Symptomatic non-parasitic hepatic cysts with biliary communication are rare and no standard treatment has been established yet. Careful attention should be paid to avoidance of postoperative bile leakage during surgical treatment.
PRESENTATION OF CASE: We report the case of a 74-year-old man who visited our department complaining of right upper abdominal pain and elevated serum levels of the liver enzymes. Computed tomography revealed hepatic cysts including a large one measuring 16 cm in diameter in Segments IV and VIII. Percutaneous drainage of the cyst revealed bile-staining of the cyst fluid. Endoscopic retrograde cholangiography demonstrated the presence of a cyst–biliary communication. We performed open deroofing of the cyst. During the operation, the biliary fistula was invisible, however, air injection into the bile duct through the stump of the cystic duct caused release of air bubbles from the cyst cavity, which allowed us to detect the small biliary orifice and repair it successfully by suture.
DISCUSSION: We utilized the intraoperative air leak test, which has previously been reported to be effective for preventing postoperative bile leakage in patients undergoing hepatectomy to detect of a small cyst–biliary communication in a case undergoing non-parasitic hepatic cyst surgery.
CONCLUSION: An intraoperative air leak test may be a useful test during surgical treatment of non-parasitic hepatic cysts with biliary communication.

1. Introduction

Symptomatic non-parasitic hepatic cysts have been treated by various methods, including percutaneous aspiration, sclerotherapy, laparoscopic or open fenestration, deroofing, cystectomy, and hepatectomy [1–4].

Cyst–biliary communication, which is difficult to diagnose by non-interventional radiological studies alone, is a rather uncommon complication that can be incidentally found during the therapeutic procedure [5–10]. Although the optimal treatment strategy for such rare cases is not established, surgical treatment may be the treatment modality of first choice because percutaneous aspiration and sclerotherapy with alcohol or minocycline, as the alternative treatment, ablates the cyst cavity and may lead to irreversible sclerosing cholangitis in cases of cysts with biliary communication [4].

If biliary communication of a hepatic cyst is demonstrated preoperatively or intraoperatively, it is important to repair the biliary fistula meticulously during the operation to avoid the postoperative bile leakage, which may increase the morbidity and postoperative length of hospital stay [11]. However, it is not always easy to identify the site of communication during the operation, especially when the size of the biliary communication is too small to detect, causing stress to the surgeons.

The trans-cystic air leak test, previously described by Zimmitti et al. was reported as a useful technique for intraoperative localization of bile leakage in hepatectomy [12].

We utilized this technique to detect the difficult-to-find intracystic biliary orifice and repaired it successfully while carrying out an open deroofing operation for a non-parasitic cyst with biliary communication.

2. Presentation of case

A 74-year-old man visited our department with the chief complaint of right upper abdominal pain. He had no fever and his blood cell counts were normal, however, the blood chemistry showed elevation of the serum C-reactive protein to 2.41 mg/dL; gamma-

Abbreviations: CT, computed tomography; US, ultrasonography; ERCP, endoscopic retrograde cholangiography.

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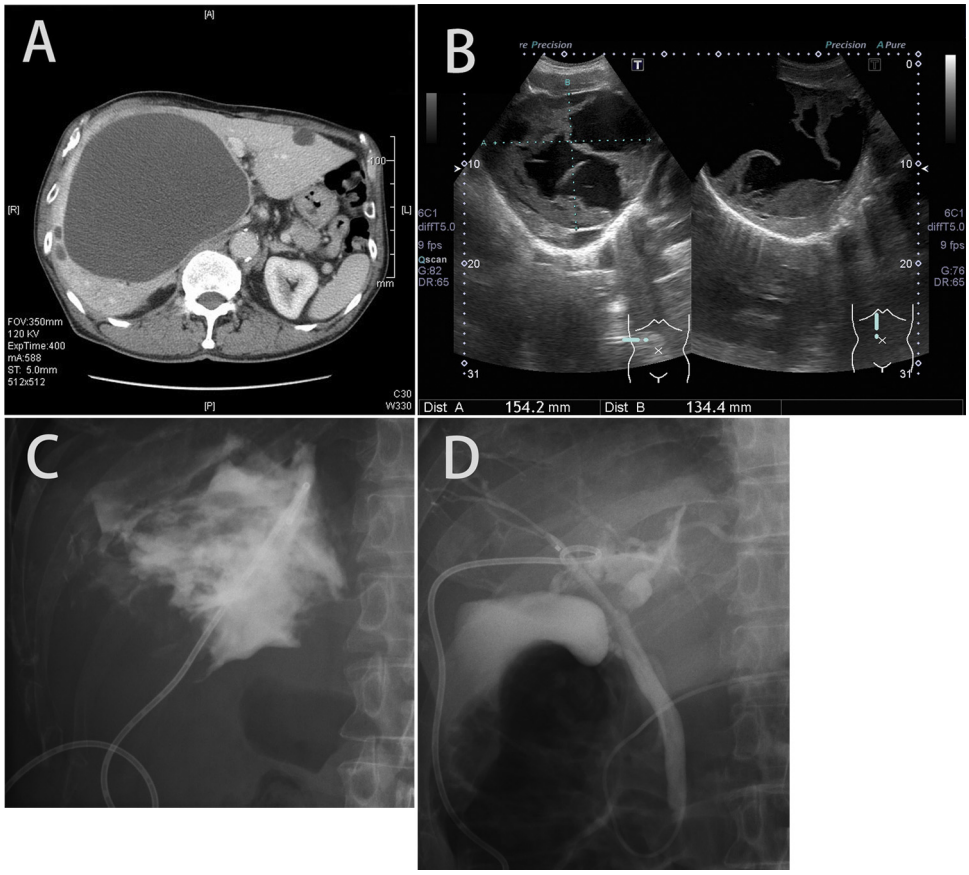

Fig. 1. Preoperative image findings.
(A) Contrast-enhanced CT showing a non-enhancing homogeneous low-density area measuring 16 cm in diameter, occupying the right paramedian sector of the liver. (B) US showing hyperechoic irregular-shaped structures in the cystic mass, suggestive of intracystic hemorrhage. (C) Cystography 5 days after percutaneous transhepatic drainage. The cyst cavity is filled with contrast material, which did not flow into the biliary tree. (D) ERCP showed the communication between the cyst and the biliary tree, but the site of communication was not clear.

glutamic transpeptitase to 510 IU/L, and alkaline phosphatase to 903 IU/L. Contrast-enhanced computed tomography (CT) of the abdomen revealed large cysts in the liver (Fig. 1A). The presence of the hepatic cysts had been pointed out during a routine CT checkup carried out at the time of endoscopic treatment of early gastric cancer 5 years earlier, however, they were left untreated because there were no symptoms. The largest cyst, located in right paramedian sector of the liver was 9 cm in diameter at the initial detection 5 years earlier, but had increased markedly in maximum diameter to 16 cm during the interventing 5-year period; it exhibited homogenous density with no mural nodules or irregular enhancement. There were no luminal septations, however, the intrahepatic biliary tree was compressed. Ultrasonography (US) revealed the large cystic mass in the right liver containing hyperechoic structures in the cyst cavity which were not visualized in enhanced CT (Fig. 1B).

We performed radiology-guided percutaneous transhepatic drainage of the cyst both for differential diagnosis from a neoplasm and for the relief of the abdominal symptoms. The discharge from the pigtail drainage catheter was initially dark-brownish colored, then changing gradually to pure bile with a total bilirubin concentration of 74.2 mg/dL. Cytological examination showed no malignant cells. Bacterial culture of the discharge fluid showed no growth. The bilious discharge from the external drainage catheter persisted at 400 mL/day for a prolonged period. While cystography conducted 5 days after the tube placement failed to reveal any evidence of biliary communication (Fig. 1C), endoscopic retrograde cholangiography (ERCP) revealed the cyst–biliary communication (Fig. 1D).

Based on the diagnosis of a non-parasitic hepatic cyst with biliary communication associated with intracystic hemorrhage, we performed a radical operation 22 days after the percutaneous drain insertion into the cyst.

At laparotomy, the large drained cyst mainly located in the right paramedian sector was found to be deep-seated beneath the thick liver parenchyma of Segments IV and VIII. The other cysts were exposed and found to be covered by a transparent capsule and to contain serous fluid. After wide resection of the dome of the cyst with covering hepatic parenchyma using a Vessel sealing system (LigaSure®, Covidien, Japan), the necrotic tissues were found in the cyst cavity and were removed as much as possible.

The inner surface of the cyst wall was examined meticulously to identify the biliary communication, but there was no visible biliary fistula. Then we performed cholecystectomy and inserted a 5 Fr silicon catheter through the stump of cystic duct, fixing it by ligation. At first, we injected radiological contrast material into the tube and performed intraoperative cholangiography. Images of the biliary tree showed the contrast material flowing into the cyst cavity from the intrahepatic bile duct, however, but the site of biliary communication could still not be clearly identified.

Then, the cyst cavity was moistened with normal saline, and air was injected into the bile duct with manual occlusion of the distal common duct; this resulted in the appearance of air bubbles inside the cyst cavity (Fig. 2), and gradual suctioning of the pooled saline and meticulous removal of the covering necrotic tissues guided by the bubbles led us to identify the hidden small biliary fistula orifice.