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Intracystic Hemorrhage with Spontaneous Rupture of Liver Cyst Complicated by Infection: A Case Report

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Abstract

A 78-year-old man who was being followed-up for a liver cyst was admitted to Asakayama General Hospital because of the sudden onset of severe right hypochondralgia and fever, with a preceding dull pain in the right upper quadrant of the abdomen of four days' duration. Computed tomography revealed remarkable enlargement of the liver cyst. We diagnosed intracystic hemorrhage and spontaneous rupture of the liver cyst. As we diagnosed bacterial infection in the ruptured cyst, transhepatic cystic drainage was performed. After improving the clinical status of the patient, we injected ethanol into the cyst, inducing a significant decrease in its size. There has been a few previous reports of intracystic hemorrhage and spontaneous rupture of a liver cyst complicated by infection. Transhepatic cyst drainage combined with antibiotic therapy and intracystic ethanol injection may be a useful and minimally invasive method for liver cysts with these complications, especially in compromised patients.

Key Words: Liver cyst; Intracystic hemorrhage; Spontaneous rupture; Infected liver cyst

Introduction

Liver cysts are not uncommon disease, occuring approximately 0.1% of the population. Benign nonparasitic liver cysts sometimes reach a large size before producing symptoms, unless complications such as rupture, bleeding, infection, and obstructive jaundice occur. However, spontaneous rupture of a liver cyst with intracystic hemorrhage is a rare complication[1-3]. We describe a patient with a liver cyst associated with infection.

Case Report

A 78-year-old man, who was being followed-up for a liver cyst after distal gastrectomy for gastric cancer 10 years ago, was admitted to Asakayama General Hospital because of the sudden

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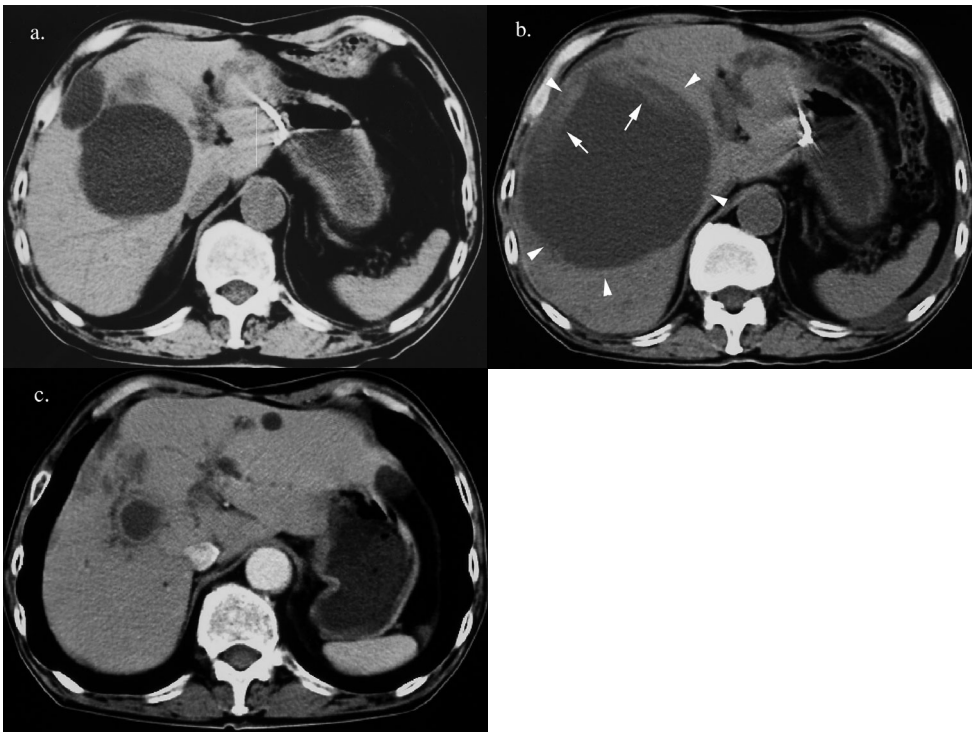

Figure 1. Computed tomograms (b) show remarkable enlargement of the liver cyst as compared with previously (a). The computed tomogram demonstrates a high density structure (arrow) in the cyst (arrowhead) and ascites including a high density area. There is no elevated lesion detected inside of the cystic wall, which indicates intracystic hemorrhage and spontaneous rupture of the liver cyst. (c) Computed tomogram four months after discharge from the hospital reveals that the cyst has decreased in size.

onset of severe right hypochondralgia and fever, with a preceding dull pain in the right upper quadrant of the abdomen of four days' duration. He had no history of chronic disease such as diabetes mellitus. Physical examination revealed spontaneous pain, and tenderness in the right hypochondral region without muscular defense. Laboratory data showed anemia (red blood cells; $257 \times 10^4/\mu L$, hemoglobin; 8.0 g/dL, hematocrit; 25.4%) and increased serum levels of C-reactive protein (28.6 mg/dL), alkaline phosphates (741 IU/L), and γ-glutamyl transpeptidase (155 IU/L). The serum concentration of bilirubin, transaminase activities and serum concentrations of tumor markers such as carcinoembrionic antigen (CEA; 1.4 ng/mL) and carbohydrate antigen 19-9 (22 U/mL) were within the reference range. Computed tomography revealed remarkable enlargement of the liver cyst in the right lobe as compared with previously (Fig. 1a), and demonstrated high density structures (arrow) in the cyst (arrowhead) and ascites including a high density area, which indicated intracystic hemorrhage and spontaneous rupture of the liver cyst (Fig. 1b). There was no elevated lesion detected inside of the cystic wall. Ultrasonography showed multiple cystic lesions with a surrounding hyperechoic area with internal septation (Fig. 2) and fluid collection between the liver and the abdominal wall. After admission, he had a continuous high-grade fever of 38℃ and right hypochondralgia. These findings strongly suggested that he was suffering from bacterial infection due to intracystic hemorrhage and spontaneous rupture of the liver cyst associated with infection. In addition to the intravenous administration of the antibiotics (Cefmetazole; 2 g/day) for seven days, we performed percutaneous transhepatic cystic drainage under ultrasonographic guidance and 250 mL volume of dark-bloody-coloured pus was

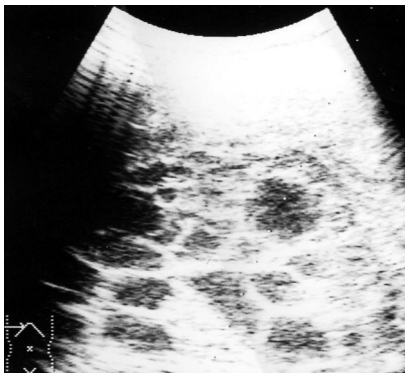

Figure 2. Ultrasonogram shows a cystic lesion with a surrounding hyperechoic area and internal septation in the right lobe of the liver, and fluid collection between the liver surface and the abdominal wall.

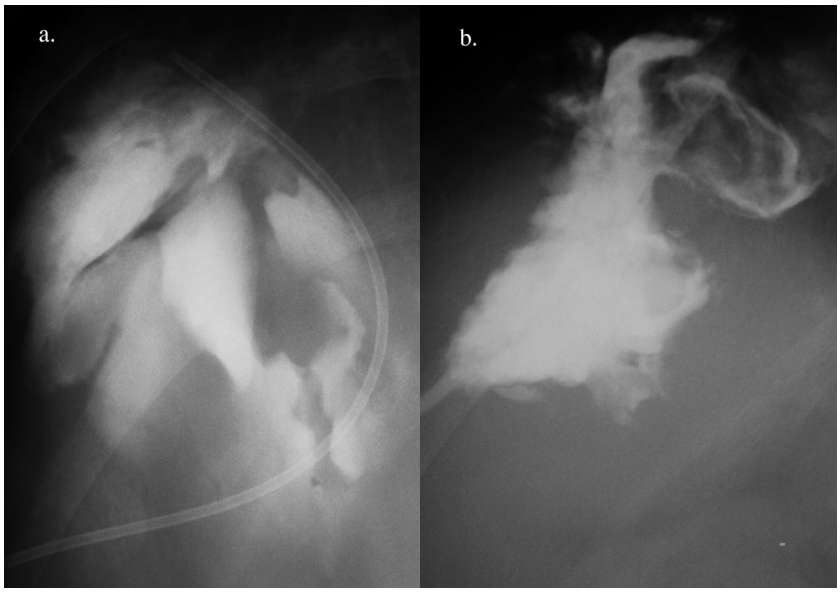

Figure 3. (a) X-ray with contrast medium via the drainage catheter on the day the cyst was drained shows a large multilobular form but no communication with biliary tract. (b) The cyst cavity has decreased in size 21 days after the drainage.

aspirated. X-ray with contrast medium via the drainage catheter showed a large multilobular form but there was no communication with the biliary tract (Fig. 3a). A culture of the fluid showed *Klebsiella pneumoniae* growth and the cytology of the fluid was negative. After the drainage, the patient recovered dramatically with decreased serum levels of C-reactive protein, alkaline phosphatase, and γ-glutamyl transpeptidase besides a transient progression of anemia which was controlled by blood transfusion. The drainage volume of the cyst decreased gradually and X-ray with contrast medium via the drainage catheter showed that the cavity had decreased in size 21 days after the drainage (Fig. 3b). Fifteen ml of 99% sterile ethanol was injected, left for 20 minutes and then removed. As the discharge persisted, a second session of ethanol sclerosis was performed seven days later. The patient was discharged from the hospital in good condition after removal of the catheter. Computed tomography four months after disharge showed that the size of the cyst had remarkably decreased (Fig. 1c).

Discussion

Intracystic hemorrhage with spontaneous rupture is an uncommon complication in nonparasitic

liver cysts[1-3]. The clinical course of this liver cyst complication usually begins with the sudden onset of severe abdominal pain. Our patient had suffered dull pain in the right upper quadrant of the abdomen for four days before the spontaneous rupture complicated with intracystic hemorrhage occurred. Enlargement of such cysts for a short period often causes abdominal pain and may play a role in the development of spontaneous rupture[4].

Although the etiology of bleeding in liver cysts is still unclear[1], rupture of the vessels in the cyst wall may be caused by rapid enlargement of the cyst and part of the cyst wall rupturing into the peritoneum. As our patient showed infection signs after abdominal pain, it was suspected that intracystic hemorrhage and spontaneous rupture of the liver cyst occurred initially and were followed by infection.

The mechanism of liver cyst infection is generally suspected to be due to gut manipulation during abdominal surgery, chronic haemodialysis, diabetes mellitus, and immunosuppressive therapy following organ transplantation, which could induce the severe depression of many defense mechanisms in the host. However, no communication with the biliary tract was demonstrated in our patient, and it may be possible that *Klebsiella pneumoniae* came from the bile; bile stasis after distal gastrectomy might cause contamination of the fluid in the cyst.

The distinction between hepatobiliary cystic neoplasm and simple hepatic cyst complicated by intracystic hemorrhage may prove difficult to determine from the clinical and radiological features because of the presence of intracystic structure in both conditions[5]. There are some reports of patients dying of sepsis without a definite diagnosis of infected cyst[6]. Surgical resection was performed in the patients with a liver cyst with intracystic hemorrhage and spontaneous rupture, because of persistent symptoms, enlargement of the cyst, and suspected malignancy[2,3]. More recently, minimally invasive therapeutic approaches have play an increasing role in the management of cystic diseases of the liver. In our patient, transhepatic cyst drainage was performed because the patient was in a septic state and was too old to undergo right lobectomy. Transhepatic cyst drainage combined with antibiotic therapy and intracystic ethanol injection, which also has a hemostatic effect via sclerosis of the bleeding vessels, may be a useful and less invasive method for high risk patients including sepsis and advanced age compared with major hepatectomy. However, it is necessary to follow-up these cases closely because of the possibility of neoplastic cysts.

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