Spontaneous Rupture of a Simple Hepatic Cyst Complicated With Intracystic Hemorrhage

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A 69-year-old female presented with abdominal pain for 3 days without trauma history. Ultrasound examination showed that she had a hepatic cystic lesion, which was enhanced on contrast computed tomography. Surgical intervention was performed 6 days after admission; bloody ascites in the peritoneal cavity, a ruptured cyst 12 cm in size, and a rupture hole over the cystic wall were noted. Spontaneous rupture is an infrequent presentation in a simple hepatic cyst with only a few case reports in the English and Japanese literature. Spontaneous rupture of a simple hepatic cyst with intracystic hemorrhage can pose a problem in differential diagnosis from cystic neoplasm of the liver and needs further management.

KEY WORDS — liver cyst, spontaneous rupture

Introduction

Most simple hepatic cysts are found incidentally. They are usually asymptomatic and do not require further treatment [1]. Complications of simple hepatic cysts include rupture, bleeding, infection and obstructive jaundice. Although these complications are rare, they require treatment upon presentation [2]. The spontaneous rupture of simple hepatic cyst is uncommon [3]. When a hepatic cyst contains lots of blood; which is a very rare occurrence, it may result in difficulty in differentiating the hepatic cyst from cystic neoplasm of the liver [1]. We present here a patient with the rare condition of spontaneous rupture of a simple hepatic cyst with internal bleeding.

Case Report

A 69-year-old woman suffering from epigastric pain and vomiting for 3 days, followed by transient loss of consciousness before admission was sent to the emergency room and then admitted to the ward of Jen-Ai General Hospital for further evaluation and management.
The patient had a history of hypertensive cardio-vascular disease with regular treatment for several years. She did not smoke or drink alcohol. The family denied any recent event that could have caused any trauma. Upon arrival at the emergency room, the patient appeared ill physically, but was conscious, lucid, and cooperative. Vital signs were as follows, blood pressure, 128/76 mmHg, pulse rate, 88 beats/min, respiratory rate, 18 breaths/min and temperature, 36.2°C. Her sclera was not icteric but conjunctiva was mildly pale. Chest examination showed clear breathing sounds over both lung fields. Observation of the heart showed a regular heartbeat without murmur. Distension of the abdomen was present but there was no significant abdominal tenderness.

Laboratory data showed that the patient’s white blood cell count was $9.050 \times 10^9$ with mild left shifting of 75%, which were in segment form. With normal ranges expressed in parentheses, hematocrit of the patient was 0.34 (0.35–0.48), hemoglobin was 110 g/L (120–160 g/L), sodium was 142 mmol/L (137–153 mmol/L), potassium was 4.0 mmol/L (3.5–5.3 mmol/L), blood urea nitrogen was 6.8 mmol/L (2.5–7.1 mmol/L), creatinine was 97.2 μmol/L (61.9–132.6 μmol/L), serum glutamic oxaloacetic transaminase was 37 U/L (10–35 U/L), serum glutamic pyruvic transaminase was 24 U/L (3–30 U/L), alkaline phosphatase was 2.3 μkat/L (0.1–4.5 μkat/L) and total bilirubin was 25.7 μmol/L (6.8–23.9 μmol/L). Tumor marker levels examined after admission, including alpha-fetoprotein (4.8 ng/mL; 0–6 ng/mL) and carbohydrate antigen 19-9 (CA19-9, 24.1 U/mL; 0–33 U/mL), were within normal limits. A chest roentgenogram showed normal lung fields and an electrocardiogram showed normal sinus rhythm.

Abdominal ultrasonogram was performed on the second day of admission. We found a huge cystic lesion approximately 13.0 × 9.5 cm in size with internal echogenicity and fluid retention in the abdominal cavity without internal septations. (Fig. 1). Computed tomography (CT) was performed later and this showed a huge cystic lesion, predominantly in the left lobe, approximately 14 cm in size with ill-defined internal high density and some fluid (Fig. 2). The differential diagnosis included complicated cyst and cystic tumor. Diagnostic tapping was performed and bloody ascites was collected. A routine examination showed a white blood cell count of 16,620 with a differential count of neutrophil/lymphocyte 86/14 and red blood cell count of 333,0000. Rupture of the cyst with bleeding into the peritoneum was suspected according to the above findings. Hemoglobin levels decreased to as low as 64 g/L and blood transfusion was given. An angiography of superior mesenteric artery was performed by a radiologist on the next day for the possibility of active bleeding, and it showed a negative finding. A general surgery doctor suggested conservative treatment initially.
Because of persistence of symptoms including abdominal pain, progressive anemia and the onset of fever on the sixth day of admission, surgical excision of the cyst was performed on the eighth day of admission. Operative findings showed approximately 1,200 mL of bloody ascites in the peritoneal cavity, a ruptured cyst approximately 12 cm in size over the left liver lobe, and a rupture hole approximately 1 cm in size over the cystic wall. Cross section of the cyst revealed a great deal of hematoma within the cyst and erosion of vessels on the cystic wall. The post operative course was uneventful and the patient was discharged 15 days after the operation. Her condition remains stable after 6 months of outpatient follow up.

Discussion

Hepatic cysts are classified as congenital, traumatic, infectious or neoplastic. Congenital hepatic cysts include simple cysts and adult polycystic liver disease. Simple hepatic cysts are usually asymptomatic and are found incidentally by abdominal ultrasonography or CT scans [4]. These cysts are detected by medical health checks. The prevalence of congenital hepatic cysts is reported to be 2–5% [5,6]. The current case was found to have had a hepatic cyst several years ago, but she did not receive regular follow up.

Complications such as hemorrhage, rupture, infection, or obstructive jaundice may occur, and require treatment when they do [2]. Hepatic cysts usually become quite large before causing any noticeable symptoms [6]. These symptoms are non-specific and include abdominal pain, abdominal discomfort and nausea [2]. Sudden onset of abdominal pain is the most frequent symptom in spontaneous rupture of simple hepatic cysts, but most cases do not present specific clinical symptoms [3]. According to Poggi et al [6], only 14 cases of rupture of a simple hepatic cyst have been reported; 13 patients had spontaneous rupture and only one patient was secondary to trauma. Enlargement of simple hepatic cysts for a short period can cause abdominal pain, and contributes to the development of spontaneous rupture [3]. The clinical course usually begins with sudden abdominal pain and is followed by an uneventful healing process with conservative treatment [3]. However, surgical treatment cannot be avoided in patients with persistent symptoms, progressive enlargement of the cyst, and suspected malignancy [3]. The present case received surgery because of the persistent symptoms of abdominal distension, pain and significant anemia.

The etiology of hemorrhage into hepatic cysts is not clear. It may be caused by vessel rupture in the cyst wall by rapid enlargement of the lesion [7]. Hemangioma and vascular malformation near or in the cyst wall or trauma may cause intracystic hemorrhage [2]. The wall of a hepatic cyst consists of three layers. The epithelial lining may become necrotic and slough if the intracystic pressure becomes too high. Injury to fragile blood vessels in the cyst wall may also induce intracystic hemorrhage [5].

Differential diagnosis of a hepatic cyst with intracystic hemorrhage with a hepatobiliary cystic neoplasm is very difficult based on clinical and radiological features because both have intracystic structures [4]. Consequently, mass hepatectomy has been unnecessarily performed for many benign hepatic cysts with intracystic hemorrhage because of the mistaken preoperative diagnosis of cystadenocarcinoma [4]. Some modalities, such as cytologic examination of cystic fluid by ultrasonic guided aspiration, or measurement of CA19-9 level may help in differential diagnosis. For example, CA19-9 levels have been reported as a possible way to differentiate hepatic cysts from cystic neoplasm of the liver because levels of this serum are higher in patients with cystic neoplasm [7]. Rupture of the cyst may raise serum CA19-9 levels by absorption of cystic fluid via the peritoneum [4]. On the other hand, CA19-9 levels might be high in cystic fluid because epithelial cells in the cyst wall express CA19-9 immunoreactivity [4]. The tumor marker levels in our patient were all within normal limits, and thus these levels do not seem to be useful for differentiation diagnosis here. Characteristic findings of cystic neoplasm on ultrasound include high
echogenic internal septations, and on CT, a thickened walled cyst with thin internal septations. Other imaging findings of cystic neoplasm may include communication between the intrahepatic bile duct and cystic lesion, and septal enhancement as a result of papillary growth along the septum. Repeated observation of the intracystic structure by CT or ultrasonography can be considered the most reliable diagnostic method for the detection of a hepatic cyst [3].

Typical ultrasonic findings of hemorrhagic hepatic cysts are cystic fluid that is hyperechoic compared with the fluid in simple cysts and internal echoes that mimic septations or solid portions [2]. Other reported findings include complete disappearance of a known cyst, a large anechoic lesion with irregular borders, homogenous echoic mass with dilatation of bile ducts, and heterogenous echoic mass with fluid retention in the abdominal cavity [6]. CT cannot clearly visualize an intracystic blood clot or hemorrhage, both of which can be defined by ultrasonography. Therefore, discrepancies between ultrasonography and CT findings are important for establishing a diagnostic practice for hepatic cysts [1]. Our findings in the present case are consistent with some of the above descriptions.

Treatment for symptomatic hepatic cysts includes percutaneous aspiration and surgery. Percutaneous aspiration with injection of minocycline hydrochloride is performed to prevent recurrence [4]. Surgery cannot be avoided in patients with persistent symptoms, progressive enlargement of size or suspicion of malignancy [5].

In conclusion, we treated a case with spontaneous rupture of a hepatic cyst with intracystic hemorrhage, which is a rare complication, with only a few cases previously reported in the English and Japanese literature [7]. Intracystic hemorrhage is also uncommon and spontaneous rupture of hepatic cyst with intracystic hemorrhage is even rarer [3]. Although most simple hepatic cysts are asymptomatic, complications of hepatic cysts may occur and differential diagnosis from cystic neoplasm should always be considered.

References