

Intracystic Hemorrhage Required No Treatment from One of Multiple Hepatic Cysts

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Abstract

We describe a patient with intracystic hemorrhage from one of multiple hepatic cysts. A 66-year-old woman was admitted to Nippon Medical School Hospital because of pain in the right upper quadrant of the abdomen. The medical history included multiple hepatic cysts and angina pectoris, which had been treated with aspirin. Three weeks before presentation, pain occurred in the right upper quadrant of the abdomen but resolved spontaneously. Ultrasonography revealed multiple hepatic cysts. One of the cysts in segment 8 had a hypoechoic structure and contained fluid. Computed tomography showed an area of homogenous density (diameter, 6 cm) which was slightly greater than that of the other hepatic cysts in segment 8. There was calcification of the cyst wall. On magnetic resonance imaging, this cyst showed heterogeneous hyperintensity on T1- and T2- weighted sequences which was greater than that of the other hepatic cysts. Intracystic hemorrhage of one of the multiple hepatic cysts was diagnosed. The pain gradually resolved without drainage, embolization, or operation, and the patient was discharged. After discharge, the upper abdominal pain did not recur. On magnetic resonance imaging 14 months later, the cyst showed heterogeneous hyperintensity on T1- and T2- weighted sequences which was less than that on the previous scan.

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Key words: intracystic hemorrhage, hepatic cyst, multiple, liver

Introduction

The prevalence of congenital hepatic cysts in the

general population is estimated to be 2% to 4%.
Most benign, nonparasitic hepatic cysts are
asymptomatic, but complications can occur.
Documented complications include obstructive

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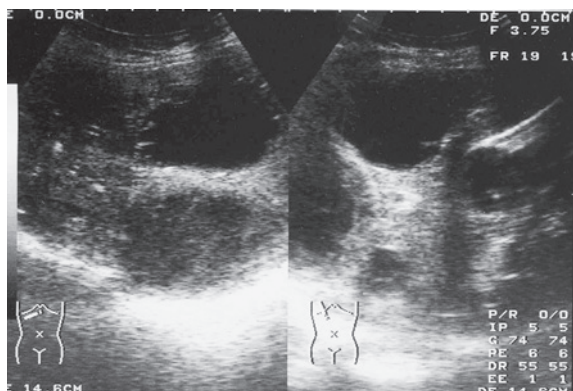


Fig. 1 An ultrasonographic image, showing multiple hepatic cysts. One of the cysts in segment 8 had a hypoechoic structure and contained fluid.

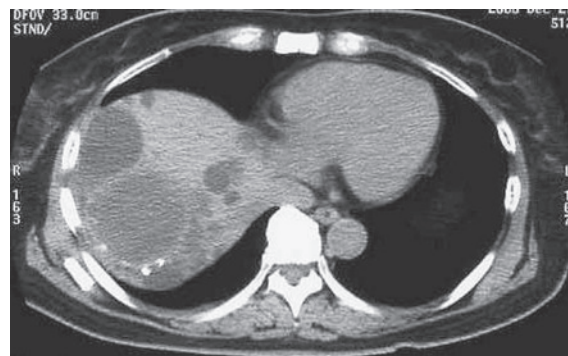


Fig. 2 A computed tomographic (CT) scan showing an area of homogenous density (diameter, 6 cm) which was slightly greater than that of the other hepatic cysts in segment 8. There was calcification of the cyst wall.

jaundice², rupture³⁻⁷, intracystic hemorrhage⁸⁻¹², and infection¹³⁻²¹; the complication rate has been reported to be about 10%, and hemorrhage and infection are the most common complications²². Therefore, intracystic hemorrhage, especially from one of multiple hepatic cysts, is a rare complication, and few cases have been reported worldwide. We describe a patient with intracystic hemorrhage from one of multiple hepatic cysts.

Case Report

A 66-year-old woman was admitted to Nippon Medical School Hospital because of pain in the right upper quadrant of the abdomen. The past medical history included multiple hepatic cysts and angina pectoris, which had been treated with aspirin. Three weeks before presentation, pain occurred in right upper quadrant of the abdomen but resolved spontaneously. The results of initial laboratory tests were as follows: serum aspartate aminotransferase, 21 IU/L (normal, <28 IU/L); serum alanine aminotransferase, 18 IU/L (normal, <33 IU/L); serum lactic dehydrogenase, 206 IU/L (normal, 180 to 460 IU/L); serum gamma glutamic transpeptidase, 19 IU/L (normal, 8 to 39 IU/L); serum C-reactive protein, 0.23 mg/dL (normal, <0.3 mg/dL); white blood cell count, 3,200 / μ L (normal, 4,000 to 8,000/ μ L); red blood cell count, 429 $\times 10^4$ / μ L (normal, 410 to 550 $\times 10^4$ / μ L); serum hemoglobin concentration, 13.4 g/dL (normal, 14 to 18 g/dL); and platelet count, 12.3 \times

10^4 / μ L (normal, 20 to 40 $\times 10^4$ / μ L). The prothrombin time was 97.5% (normal, 70% to 130%), and the activated partial thromboplastin time was 26.3 seconds (normal, 24 to 37 seconds). Ultrasonography revealed multiple hepatic cysts. One of the cysts in segment 8 had a hypoechoic structure and contained fluid (**Fig. 1**). Computed tomography (CT) showed an area of homogenous density (diameter, 6 cm) which was slightly greater than that of the other hepatic cysts in segment 8. The cyst wall was calcified (**Fig. 2**). On magnetic resonance imaging (MRI), this cyst showed heterogeneous hyperintensity on T1- and T2- weighted sequences which was greater than that of the other hepatic cysts (**Fig. 3a, b**). Intracystic hemorrhage of one of the multiple hepatic cysts was diagnosed. Pain gradually resolved without drainage, embolization, or operation, and the patient was discharged. After discharge, the abdominal pain did not recur. On MRI 14 months later, the cyst showed heterogeneous hyperintensity on T1- and T2- weighted sequences which was less than that on the previous MRI scan (**Fig. 3c, d**).

Discussion

Simple hepatic cysts are common, benign, usually asymptomatic, and require no treatment. They occasionally resolve spontaneously but can also cause serious complications^{2-19,21}. Symptoms of hemorrhagic hepatic cysts are nonspecific and include abdominal pain, discomfort, and nausea. In

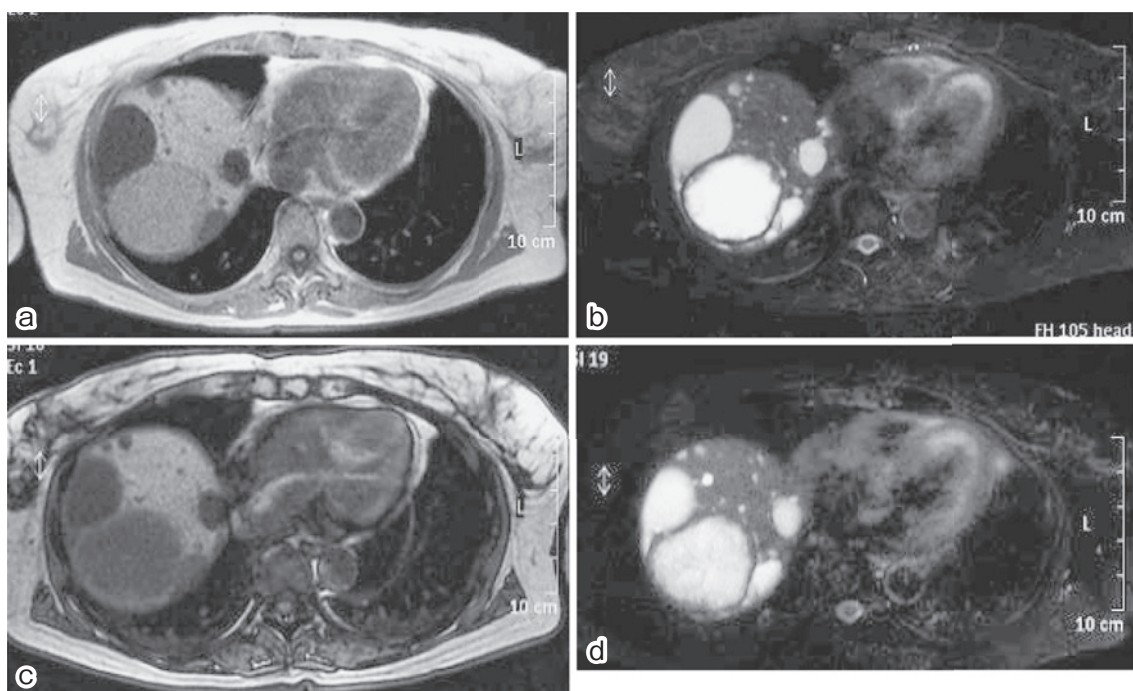


Fig. 3 On magnetic resonance imaging (MRI), this cyst showed heterogeneous hyperintensity on T1- (a) and T2- (b) weighted sequences greater than that of the other hepatic cysts. On magnetic resonance imaging (MRI) 14 months later, the cyst showed heterogeneous hyperintensity on T1- (c) and T2- (d) weighted sequences which was less than that on the previous MRI scan.

most cases, the cause of the intracystic hemorrhage is unclear. The wall of a hepatic cyst consists of 3 layers: an inner layer of loose connective tissue lined with cylindrical or cuboidal epithelium, a middle layer of compact connective tissue containing blood vessels, and an outer layer of loose connective tissue with large blood vessels, bile ducts, and occasional von Meyenburg complexes. The epithelial lining may undergo necrosis and sloughing if the intracystic pressure becomes too high. Injury to fragile blood vessels in the cyst wall and an increased risk of bleeding associated with the use of aspirin may thus have been responsible for the intracystic hemorrhage in our patient²³.

Biliary cystadenocarcinomas are rare cystic tumors arising from the biliary epithelium. Differentiating hepatobiliary cystic neoplasms from simple hepatic cysts with internal hemorrhage is often difficult on the basis of clinical and radiological features because both lesions have intracystic structures^{11,24}. However, some diagnostic techniques are useful for differentiating hemorrhagic hepatic cysts from cystic neoplasms^{9,24-27}. Cytologic examination of cystic fluid obtained with aspiration

under ultrasonographic guidance may provide important information for distinguishing malignant from benign lesions⁹. However, benign lesions are often difficult to distinguish from malignant lesions that lack characteristic features of malignancy. Moreover, aspiration of a cystic lesion can lead to tumor spread, cyst rupture, or bleeding. Ultrasonography and CT may show abnormal findings that mimic those of other conditions^{9,28}. On ultrasonographic examination, hemorrhagic hepatic cysts typically contain fluid that is hyperechogenic compared with the fluid in simple cysts. On MRI, intracystic hemorrhage is frequently associated with internal signal intensities that mimic septations or solid portions as confirmed with surgery or percutaneous aspiration, suggesting that hyperintensity on T1- and T2- weighted MRI sequences can help differentiate internally hemorrhagic cysts from other cystic lesions. On T1-weighted images, the signal intensity of fluid changes from low to high as the protein concentration increases²⁵. In our patient, the hemorrhagic cyst heterogeneously showed hyperintensity on T1- and T2- weighted sequences

which was greater than that of other hepatic cysts. After 14 months, the cyst showed heterogeneous hyperintensity on T1- and T2- weighted sequences which was less than that on the previous MRI scan. The signal intensity of hemorrhage decreases when clots are liquefied²⁹. On CT and MRI, the walls of most cysts are thin and smooth and are not enhanced after intravenous injection of contrast medium. However, enhanced thick walls are occasionally seen when inflammation, granulation, or fibrosis occurs. Calcification of the cyst wall may occur but may also be seen in simple cysts. After hemorrhage, cyst fluid is usually hyperdense on CT and T1- weighted MR images and hypointense to hyperintense on T2- weighted images. A fluid-fluid level is sometimes observed.

Treatment recommendations for symptomatic hepatic cysts include surgery³⁰⁻³² and the injection of a sclerosing agent into the cyst³³⁻³⁵. The clinical presentation of spontaneous intracystic hemorrhage usually begins with severe abdominal pain of sudden onset, followed by a gradual decrease in pain and healing with conservative therapy^{8,10,11,28}. Then, the intracystic hemorrhage may be treated conservatively and closely observed, if no acute abdominal symptoms develop⁸, and if cystic liver tumors, such as cystadenoma and cystadenocarcinoma, are ruled out with imaging studies, such as ultrasound, contrast-enhanced ultrasound³⁶, CT, and MRI.

References

- Gaines PA, Sampson MA: The prevalence and characterization of simple hepatic cysts by ultrasound examination. *Br J Radiol* 1989; 62: 335-337.
- Schwed DA, Edoga JK, Stein LB: Biliary obstruction due to spontaneous hemorrhage into benign hepatic cyst. *J Clin Gastroenterol* 1993; 16: 84-86.
- Andersson R, Tranberg KG, Bengmark S: Hemoperitoneum after spontaneous rupture of liver tumor: results of surgical treatment. *HPB Surg* 1988; 1: 81-83.
- Klingler PJ, Bodner E, Schwelberger HG: Late complication after laparoscopic fenestration of a liver cyst. *Surg Laparosc Endosc* 1998; 8: 76-77.
- Lotz GW, Stahlschmidt M: Intra-abdominal bleeding after rupture of hepatic cyst. *South Med J* 1989; 82: 667.
- Ueda J, Yoshida H, Taniai N, Mineta S, Kawano Y, Uchida E: A case of spontaneous rupture of a simple hepatic cyst. *J Nippon Med Sch* 2010; 77: 181-185.
- Yamaguchi M, Kuzume M, Matsumoto T, Matsumiya A, Nakano H, Kumada K: Spontaneous rupture of a nonparasitic liver cyst complicated by intracystic hemorrhage. *J Gastroenterol* 1999; 34: 645-648.
- Zanen AL, van Tilburg AJ: Bleeding into a liver cyst can be treated conservatively. *Eur J Gastroenterol Hepatol* 1995; 7: 91-93.
- Hanazaki K, Wakabayashi M, Mori H, et al: Hemorrhage into a simple liver cyst: diagnostic implications of a recent case. *J Gastroenterol* 1997; 32: 848-851.
- Yoshida H, Onda M, Tajiri T, et al: Intracystic hemorrhage of a simple hepatic cyst. *Hepatogastroenterology* 2002; 49: 1095-1097.
- Takahashi G, Yoshida H, Mamada Y, Taniai N, Bando K, Tajiri T: Intracystic hemorrhage of a large simple hepatic cyst. *J Nippon Med Sch* 2008; 75: 302-305.
- Yoshida H, Onda M, Tajiri T, et al: Infected hepatic cyst. *Hepatogastroenterology* 2003; 50: 507-509.
- Yoshida H, Tajiri T, Mamada Y, et al: Infected solitary hepatic cyst. *J Nippon Med Sch* 2003; 70: 515-518.
- Quigley M, Joglekar VM, Keating J, Jagath S: Fatal *Clostridium perfringens* infection of a liver cyst. *J Infect* 2003; 47: 248-250.
- Egbuna O, Johnson S, Pavlakis M: Rupture of an infected liver cyst into the pericardium in a kidney transplant recipient with polycystic kidney disease. *Am J Kidney Dis* 2007; 49: 851-853.
- Keven K, Bengisun JS, Altuntas F, et al: Cystic infection of the liver in a maintenance haemodialysis patient. *Nephrol Dial Transplant* 2001; 16: 859-860.
- Eloubeidi MA, Rockey DC: Infected hepatic cyst masquerading as abdominal aortic aneurysm. *Dig Dis Sci* 2000; 45: 1977-1980.
- Bourgeois N, Kinnart P, Vereerstraeten P, Schoutens A, Toussaint C: Infection of hepatic cysts following kidney transplantation in polycystic disease. *World J Surg* 1983; 7: 629-631.
- Shoji F, Kitamura M, Shirabe K, Kai H, Akiyoshi T, Sugimachi K: Infected hepatic cyst in a patient with multiple hepatic cysts: report of a case diagnosed by change of ultrasonographic findings. *Eur J Gastroenterol Hepatol* 2000; 12: 703-705.
- Ishii K, Yoshida H, Taniai N, Moneta S, Kawano Y, Tajiri T: Infected hepatic cyst treated with percutaneous transhepatic drainage. *J Nippon Med Sch* 2009; 76: 160-164.
- Yoshida H, Onda M, Tajiri T, et al: Spontaneous disappearance of a hepatic cyst. *J Nippon Med Sch* 2001; 68: 58-60.
- Murphy BJ, Casillas J, Ros PR, et al: The CT appearance of cystic masses of the liver. *Radiographics* 1989; 9: 307-322.
- Gaviser D: Solitary nonparasitic cysts of the liver. *Minn Med* 1953; 36: 831-836; passim.
- Horsmans Y, Laka A, Gigot JF, Geubel AP: Serum and cystic fluid CA 19-9 determinations as a diagnostic help in liver cysts of uncertain nature. *Liver* 1996; 16: 255-257.
- Vilgrain V, Silbermann O, Benhamou JP, Nahum H:

- MR imaging in intracystic hemorrhage of simple hepatic cysts. *Abdom Imaging* 1993; 18: 164–167.
26. Yoshida H, Tajiri T, Mamada Y, et al: Rapidly enlarging hepatobiliary cystadenoma. *J Med Ultrasonics* 2003; 30: 257–262.
 27. Tani A, Yoshida H, Mamada Y, Tani N, Naito Z, Tajiri T: A case of biliary cystadenocarcinoma with intracystic bleeding. *J Nippon Med Sch* 2008; 75: 293–297.
 28. Chang SS, Chan CC, Wang SS, Chang FY, Lee SD: Repeated episodes of spontaneous intracystic hemorrhage of hepatic cysts mimicking malignancy. *Zhonghua Yi Xue Za Zhi (Taipei)* 2000; 63: 256–261.
 29. Gomori JM, Grossman RI: Head and neck hemorrhage. *Magn Reson Annu* 1987; 71–112.
 30. Jones WL, Mountain JC, Warren KW: Symptomatic non-parasitic cysts of the liver. *Br J Surg* 1974; 61: 118–123.
 31. Diez J, Decoud J, Gutierrez L, Suhl A, Merello J: Laparoscopic treatment of symptomatic cysts of the liver. *Br J Surg* 1998; 85: 25–27.
 32. Klingler PJ, Gadenstatter M, Schmid T, Bodner E, Schwelberger HG: Treatment of hepatic cysts in the era of laparoscopic surgery. *Br J Surg* 1997; 84: 438–444.
 33. Yoshida H, Egami K, Onda M, Tajiri T, Uchida E: Treatment of symptomatic hepatic cyst by injection of minocycline hydrochloride. *J Hepatobiliary Pancreat Surg* 1996; 3: 491–494.
 34. Yoshida H, Onda M, Tajiri T, et al: Long-term results of multiple minocycline hydrochloride injections for the treatment of symptomatic solitary hepatic cyst. *J Gastroenterol Hepatol* 2003; 18: 595–598.
 35. Bean WJ, Rodan BA: Hepatic cysts: treatment with alcohol. *AJR Am J Roentgenol* 1985; 144: 237–241.
 36. Akiyama T, Inamori M, Saito S, et al: Levovist ultrasonography imaging in intracystic hemorrhage of simple liver cyst. *World J Gastroenterol* 2008; 14: 805–807.

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