

Intracystic Hemorrhage of a Large Simple Hepatic Cyst

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Abstract

Spontaneous intracystic hemorrhage rarely occurs in nonparasitic hepatic cysts. We describe a patient with spontaneous intracystic hemorrhage of a large simple hepatic cyst that mimicked a malignancy. A 59-year-old man presented with right abdominal discomfort. The patient's medical history included a simple hepatic cyst that had been detected 7 years earlier but was left untreated. Three weeks before presentation, right upper abdominal pain occurred but resolved spontaneously. The serum CA19-9 concentration was 48.3 U/mL (normal <37 U/mL). Ultrasonography revealed a large cystic mass, containing many hyperechoic structures and occupying nearly the entire right hepatic lobe. Computed tomography demonstrated a homogenous low-density area, 20 cm in diameter, in the right hepatic lobe. Magnetic resonance imaging revealed a heterogeneous hypointense lesion measuring 20 cm in diameter. The lesions showed linear hyperintense areas on T1-weighted sequences and mosaic heterogeneous hyperintensity on T2-weighted sequences. We suspected a hemorrhagic simple hepatic cyst, hydatid cyst, or hemorrhagic cystadenocarcinoma. Right hepatectomy was performed, and the enlarged right lobe was removed. The mass was soft, sponge-like, and contained fluid, but was not elevated. Pathologic examination of the surgical specimen confirmed the presence of a hemorrhagic benign hepatic cyst.

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Key words: intracystic hemorrhage, hepatic cyst

Introduction

Most nonparasitic hepatic cysts are asymptomatic and are found incidentally. Spontaneous intracystic hemorrhage rarely occurs in nonparasitic hepatic cysts¹⁻⁵. Occasionally, intracystic hemorrhage and cystadenocarcinoma of the liver cannot be reliably differentiated on imaging studies. Hepatectomy is usually performed in this situation²⁴. We describe a patient with spontaneous intracystic hemorrhage of

a large simple hepatic cyst that mimicked a malignancy.

Case Report

A 59-year-old man presented with right abdominal discomfort. The patient's medical history included a simple hepatic cyst that had been detected 7 years earlier but was left untreated. Three weeks before presentation, right upper abdominal pain occurred but resolved spontaneously. Results of initial

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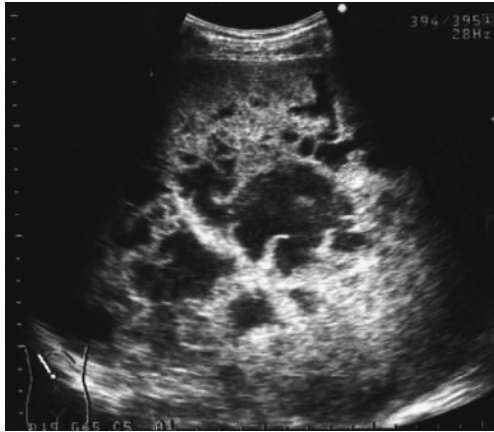


Fig. 1 Ultrasonography revealed a large cystic mass containing many hyperechoic structures and occupying nearly the entire right hepatic lobe.



Fig. 2 CT demonstrated a homogenous low-density area, 20 cm in diameter, in the right hepatic lobe. However, the structures detected on ultrasonography were not clearly visualized, even with enhanced CT.

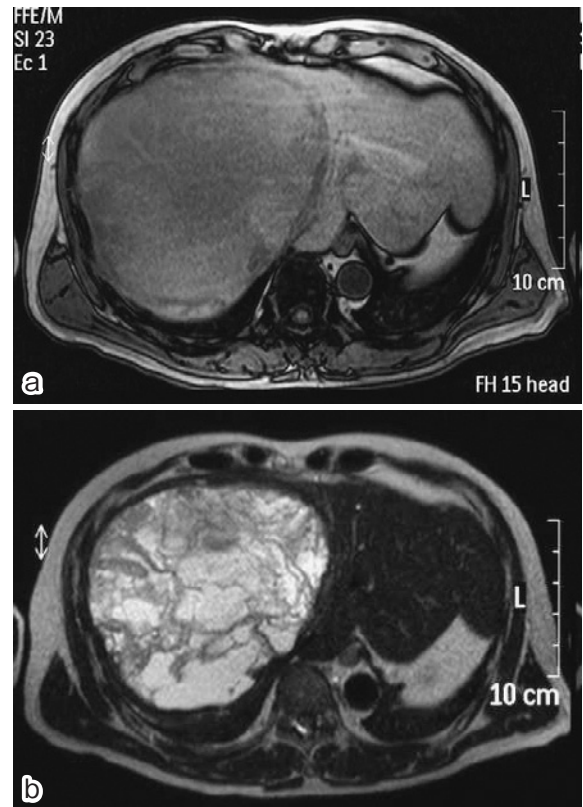


Fig. 3 MRI revealed a heterogeneous hypointense lesion 20 cm in diameter with linear hyperintense areas on T1-weighted sequences (a) and mosaic heterogeneous hyperintensity on T2-weighted sequences (b).

laboratory tests were as follows: serum aspartate aminotransferase, 21 IU/L (normal, <28 IU/L); serum alanine aminotransferase, 10 IU/L (normal, <33 IU/L); serum alkaline phosphatase, 408 IU/L (normal 66 to 220 IU/L); serum lactic dehydrogenase, 210 IU/L (normal, 180 to 460 IU/L); serum gamma glutamic transpeptidase, 324 IU/L (normal, 8 to 39 IU/L); serum C-reactive protein, 1.79 mg/dL (normal, <0.3 mg/dL); white blood cell count, 5,500 / μ L (normal, 4,000 to 8,000 / μ L); red blood cell count, 422×10^4 / μ L (normal, 410 to 550×10^4 / μ L); and serum hemoglobin concentration, 13.2 g/dL (normal, 14 to

18 g/dL). The serum concentration of carcinoembryonic antigen was 1.0 ng/mL (normal < 2.5 ng/mL), that of CA19-9 was 48.3 u/mL (normal < 37), and that of AFP was 5.6 ng/mL (normal <10). Ultrasonography revealed a large cystic mass, containing many hyperechoic structures and occupying nearly the entire right hepatic lobe (**Fig. 1**). Computed tomography (CT) demonstrated a homogenous low-density area, 20 cm in diameter, in the right hepatic lobe. However, the structures detected on ultrasonography were not clearly visualized, even with enhanced CT (**Fig. 2**). Magnetic resonance imaging (MRI) revealed a heterogeneous hypointense lesion 20 cm in diameter with linear hyperintense areas on T1-weighted sequences and mosaic heterogeneous hyperintensity on T2-weighted sequences (**Fig. 3**). We suspected a hemorrhagic simple hepatic cyst, hydatid cyst, or hemorrhagic cystadenocarcinoma.

Right hepatectomy was performed, and the



Fig. 4 The mass was soft, sponge-like, and contained fluid but was not elevated.

enlarged right lobe was removed. The mass was soft, sponge-like, and contained fluid, but was not elevated (**Fig. 4**). Pathologic examination of the surgical specimen confirmed the presence of a hemorrhagic benign hepatic cyst. The postoperative course was uneventful, and the patient was discharged 8 days after the operation.

Discussion

Most hepatic cysts are asymptomatic, but such complications as obstructive jaundice⁶, rupture^{2,7-9}, infection^{10,11}, and intracystic hemorrhage can occur¹⁻⁵. The prevalence of congenital hepatic cysts in the general population is reported to be 2% to 4%¹². Gavisser¹³ reported that the wall of a hepatic cyst consists of three 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 may thus have been responsible for the intracystic hemorrhage in our patient.

Treatment recommendations for symptomatic hepatic cysts include surgery¹⁴⁻¹⁶ and the injection of a sclerosing agent into the cyst¹⁷⁻¹⁹. The clinical presentation of spontaneous intracystic hemorrhage usually begins with severe abdominal pain of sudden onset and is followed by a gradual decline in pain

and healing with conservative therapy^{1,3,5}.

Differentiating between hepatobiliary cystic neoplasms and simple hepatic cysts complicated by intracystic hemorrhage is often difficult on the basis of clinical and radiological features because both lesions have intracystic structures²⁰. However, some diagnostic techniques can be used to differentiate hemorrhagic hepatic cysts and cystic neoplasms^{4,20-22}. Cytologic examination of cystic fluid obtained by aspiration under ultrasonographic guidance may provide important information for distinguishing malignant from benign lesions⁴. However, a benign lesion cannot be completely differentiated from malignant disease that lacks characteristic features of malignancy. Furthermore, aspiration of a cystic lesion may lead to tumor spread or to cyst rupture or bleeding. Ultrasonography and CT may show abnormal findings that mimic those of other conditions^{1,4}. Vilgrain et al²¹ have reported MRI findings in patients with intracystic hemorrhage as confirmed by surgery or percutaneous aspiration and have suggested that hyperintensity on T1- and T2-weighted MRI sequences may help to differentiate intracystic hemorrhage from other cystic lesions. On T1-weighted images, the signal intensity of fluid changes from low to high as the protein concentration increases. In our patient, the cyst showed heterogeneous hypointensity with linear hyperintense areas on T1-weighted sequences and mosaic heterogeneous hyperintensity on T2-weighted sequences. The signal intensity of hemorrhage decreases when clots are liquefied²³. In the present case, right upper abdominal pain developed 3 weeks before presentation, and MRI was performed 4 weeks after presentation. Hemorrhage of the hepatic cyst might have occurred 7 weeks before MRI. Clots might have liquefied for 7 weeks.

Levels of CA19-9 are high in the cystic fluid because epithelial cells in the cyst wall express CA19-9²⁰. Measuring concentrations of CA19-9 in serum and in the cyst fluid may help to distinguish a hemorrhagic simple cyst from a cystadenoma or cystadenocarcinoma^{22,23}. Serum levels of CA19-9 in patients with cystic neoplasms are abnormally high, whereas serum CA19-9 levels in patients with simple

hemorrhagic cyst are within the normal range²⁰. However, in our patient, the serum CA19-9 level (48.3 μ /mL) was slightly elevated. The presence of intracystic structures made it difficult to distinguish between a hepatobiliary cystic neoplasm and a hemorrhagic simple hepatic cyst. Surgery cannot be avoided in patients with suspected malignancy^{2,4}.

References

1. 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.
2. 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.
3. Zanen AL, van Tilburg AJ: Bleeding into a liver cyst can be treated conservatively. *Eur J Gastroenterol Hepatol* 1995; 7: 91-93.
4. 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.
5. Yoshida H, Onda M, Tajiri T, et al: Intracystic hemorrhage of a simple hepatic cyst. *Hepatogastroenterology* 2002; 49: 1095-1097.
6. Schwed DA, Edoga JK, Stein LB: Biliary obstruction due to spontaneous hemorrhage into benign hepatic cyst. *J Clin Gastroenterol* 1993; 16: 84-86.
7. Lotz GW, Stahlschmidt M: Intra-abdominal bleeding after rupture of hepatic cyst. *South Med J* 1989; 82: 667.
8. Andersson R, Tranberg KG, Bengmark S: Hemoperitoneum after spontaneous rupture of liver tumor: results of surgical treatment. *HPB Surg* 1988; 1: 81-83.
9. Klingler PJ, Bodner E, Schwelberger HG: Late complication after laparoscopic fenestration of a liver cyst. *Surg Laparosc Endosc* 1998; 8: 76-77.
10. Yoshida H, Onda M, Tajiri T, et al: Infected hepatic cyst. *Hepatogastroenterology* 2003; 50: 507-509.
11. Yoshida H, Tajiri T, Mamada Y, et al: Infected solitary hepatic cyst. *J Nippon Med Sch* 2003; 70: 515-518.
12. Gaines PA, Sampson MA: The prevalence and characterization of simple hepatic cysts by ultrasound examination. *Br J Radiol* 1989; 62: 335-337.
13. Gavisser D: Solitary nonparasitic cysts of the liver. *Minn Med* 1953; 36: 831-836; passim.
14. Jones WL, Mountain JC, Warren KW: Symptomatic non-parasitic cysts of the liver. *Br J Surg* 1974; 61: 118-123.
15. 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.
16. 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.
17. 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.
18. 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.
19. Bean WJ, Rodan BA: Hepatic cysts: treatment with alcohol. *AJR Am J Roentgenol* 1985; 144: 237-241.
20. Horsmans Y, Laka A, Gigot JF, Geubel AP: Serum and cystic fluid CA19-9 determinations as a diagnostic help in liver cysts of uncertain nature. *Liver* 1996; 16: 255-257.
21. Vilgrain V, Silbermann O, Benhamou JP, Nahum H: MR imaging in intracystic hemorrhage of simple hepatic cysts. *Abdom Imaging* 1993; 18: 164-167.
22. Yoshida H, Tajiri T, Mamada Y, et al: Rapidly enlarging hepatobiliary cystadenoma. *J Med Ultrasonics* 2003; 30: 257-262.
23. Gomori JM, Grossman RI: Head and neck hemorrhage. *Magn Reson Annu* 1987; 71-112.

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