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Tomoki Nakajima, Manabu Okajima, Akiko Shibuya, Junko Yamaoka, Toshiaki Nakashima, Yoshito Itoh

ABSTRACT

Introduction: Spontaneous rupture of a simple hepatic cyst is rare, and the clinical course after rupture is not fully known. We report a case of a huge hepatic cyst which rapidly regrew after spontaneous rupture.

Case Report: A 74-year-old male underwent further examination and follow-up of a huge hepatic cyst which was first detected at annual health check. On his first visit, the cyst was 10×9 cm in size. During the first 7 years of follow-up, the cyst gradually grew to 18×14 cm. Eight years after his first visit, ultrasonography showed that the huge hepatic cyst was without intracystic echogenic content. However, nine days after that, a computed tomography (CT) scan revealed that the cyst had nearly disappeared with remnant minimal cystic fluid and ascites although the patient was asymptomatic. Subsequently, within one month, the patient complained of back pain, and another CT scan showed that the cyst rapidly regrew to 13×10 cm in size. The cyst gradually regressed after sequential intracystic injection of absolute ethanol and minocycline.

Conclusion: The rapid disappearance of the cyst was considered to be due to intraperitoneal rupture. The patient was asymptomatic just after rupture, but complained of back pain possibly because the regrowth was rapid. In some cases of huge hepatic cyst, rupture may be dismissed because it can be asymptomatic and the cystic fluid may have possibly already accumulated by the time symptoms appear.



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CASE REPORT

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Introduction: Spontaneous rupture of a simple hepatic cyst is rare, and the clinical course after rupture is not fully known. We report a case of a huge hepatic cyst which rapidly regrew after spontaneous rupture. **Case Report:** A 74-year-old male underwent further examination and follow-up of a huge hepatic cyst which was first detected at annual health check. On his first visit, the cyst was 10×9 cm in size. During the first 7 years of follow-up, the cyst gradually grew to 18×14 cm. Eight years after his first visit, ultrasonography showed that the huge hepatic cyst was without intracystic echogenic content. However, nine days after that, a computed tomography (CT) scan revealed that the cyst had nearly disappeared with remnant minimal cystic fluid and ascites although the patient was asymptomatic. Subsequently, within one month, the patient complained of back pain, and another CT scan showed that the cyst rapidly regrew to 13×10 cm in size. The cyst gradually regressed after sequential intracystic injection of absolute ethanol and minocycline. **Conclusion:** The rapid disappearance of the cyst was considered to be due to intraperitoneal

rupture. The patient was asymptomatic just after rupture, but complained of back pain possibly because the regrowth was rapid. In some cases of huge hepatic cyst, rupture may be dismissed because it can be asymptomatic and the cystic fluid may have possibly already accumulated by the time symptoms appear.

Keywords: Spontaneous rupture, Hepatic cyst, Absolute ethanol, Minocycline

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INTRODUCTION

Simple hepatic cysts are considered to be congenital. They are generally stable in size over time and require no treatment [1]. But some cysts may slowly enlarge and occasionally become symptomatic due to mass effect, rupture, hemorrhage, or infection [1]. Spontaneous rupture of a simple hepatic cyst is a rare occurrence [1, 2], and the clinical course after rupture is not fully known. We report a case of a huge hepatic cyst which rapidly regrew after spontaneous rupture. To our knowledge, this is the first case report which demonstrated such a rapid change in the size and morphology of the hepatic cyst during a short period after spontaneous rupture.

CASE REPORT

A 74-year-old male visited our outpatient clinic on August 21, 2001 for further examination of a cystic lesion

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in the anterior lobe of the liver which was detected at annual health check. Magnetic resonance imaging (MRI) scan suggested that the lesion was a simple cyst of 10×9 cm in size and had no sign of infection, hemorrhage or malignancy (Figure 1). On April 30, 2008, he revisited our clinic for follow-up. A computed tomography (CT) scan demonstrated that the cyst had increased to 18×14 cm in size, but there was no evidence of complication or malignancy (Figure 2). On June 10, 2009, he visited our clinic again for follow-up of this change in the lesion. Ultrasonography showed that the huge hepatic cyst was without intracystic echogenic content (Figure 3). However, a CT scan performed on June 19, 2009 revealed that the huge cyst had nearly disappeared during this nine-day period, and only a small amount of cystic fluid remained (Figure 4A–B). Intestinal loops were seen in the region previously occupied by the large cyst. A small amount of ascites was found in contact with the anterior surface of the liver. Since the remnant cyst fluid in the cyst cavity and ascites were both minimal, the communication between these fluids was not directly shown on CT images. However, it was considered that the cyst had ruptured into the peritoneal space first, that the extravasated fluid had been mostly absorbed, and that the remnant fluid was detected as minimal ascites. There was no finding of communication between cyst cavity and biliary trees. On July 15, 2009 the patient complained of back pain, and another CT scan showed reappearance of the cyst which had grown to 13×10 cm in size (Figure 5). It was speculated that the ruptured lesion was repaired, and cystic fluid had accumulated in less than one month. The cystic fluid was macroscopically serous, and the cytology showed no sign of hemorrhage, malignancy or parasitic infection. After complete aspiration of the cystic fluid, we intracystically injected contrast medium and ensured there was no communication with the biliary tree or extravasation into the peritoneal cavity. Then, 70 mL of absolute ethanol was injected into the cyst. The ethanol was left in place for 20 minutes and all was withdrawn, as reported previously [3, 4]. One week after the injection, the cyst reappeared, thus, the therapeutic intervention was not sufficient. After complete aspiration of cystic fluid once again, 200 mg of minocycline hydrochloride dissolved in 20 mL of saline was injected into the cyst. The solution was left in place for 15 minutes and it was withdrawn [4]. Because the cyst remained for 2 months, 100 mL of absolute ethanol followed by 200 mg of minocycline hydrochloride dissolved in 20 mL of saline were sequentially injected with an interval of one week. All the intracystic injections were given under ultrasound guidance and percutaneously. Thereafter, the cyst gradually regressed; the size of the cyst was 9.0×7.3 cm on December 2, 2009, 6.7×4.8 cm on April 7, 2010, 5.9×4.3 cm on August 4, 2010, 5.9×3.0 cm on January, 2011, 3.5×2.3 cm on December 9, 2011, and 3.0×2.3 cm in the last follow-up on March 22, 2013. The liver function tests were within normal range and stable throughout this clinical course.

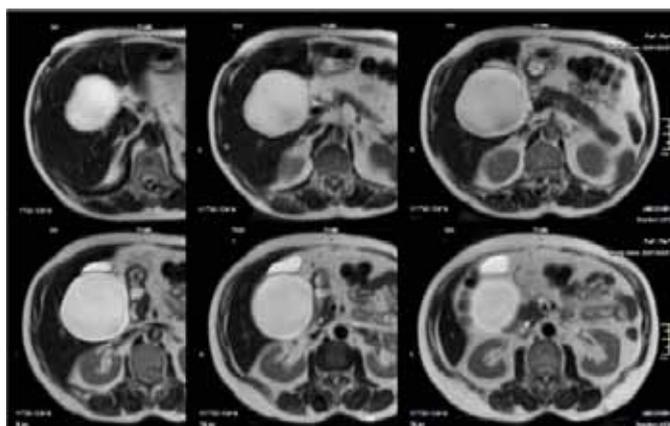


Figure 1: On August 21, 2001 magnetic resonance T2-weighted images suggested that the huge hepatic lesion was a simple cyst of 10×9 cm in size, was located in the anterior lobe and had no sign of infection, hemorrhage or malignancy.

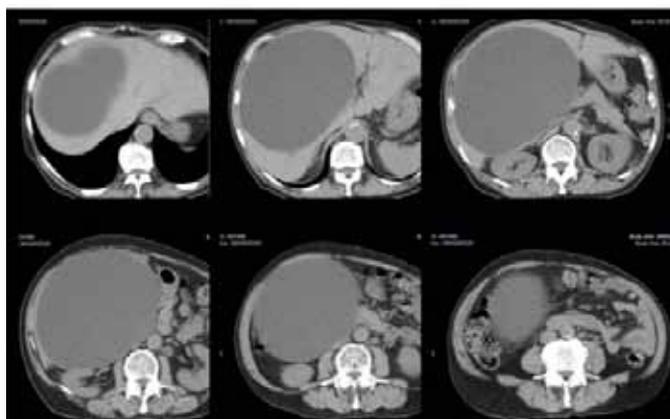


Figure 2: On April 30, 2008 a computed tomography scan demonstrated that the cyst increased to 18×14 cm in size, but there was no evidence of complication or malignancy.

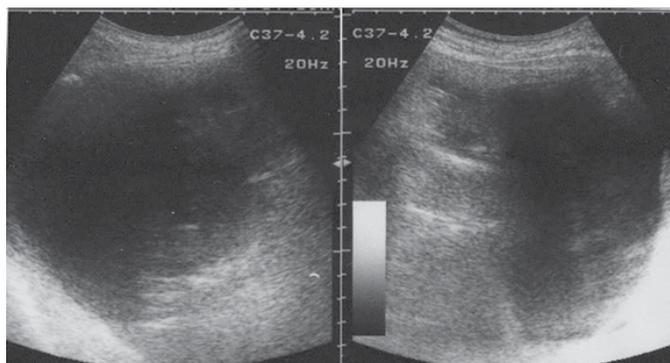


Figure 3: On June 10, 2009 ultrasonography showed that the huge hepatic cyst was without intracystic echogenic content.

DISCUSSION

There are some reports on the disappearance of non-parasitic hepatic cysts caused by intraperitoneal rupture after blunt trauma to the abdomen or coughing episodes

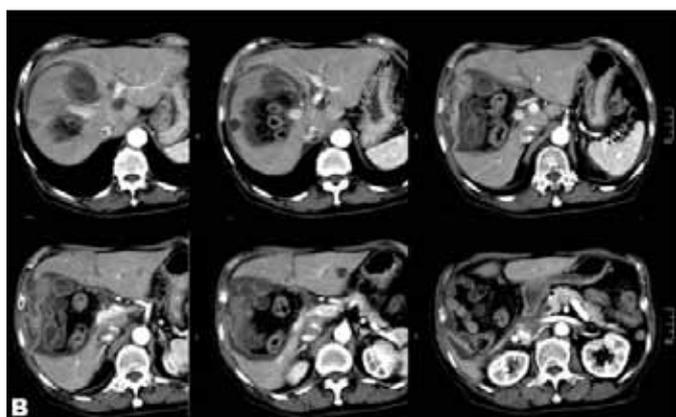
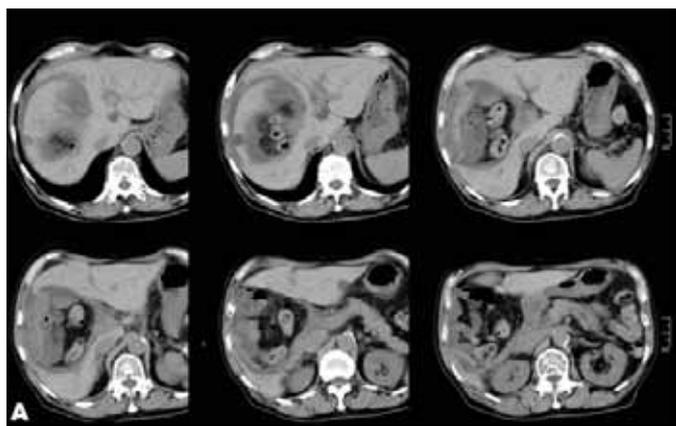


Figure 4: The computed tomography scan on June 19, 2009 revealed that the huge cyst disappeared during this nine-day period, and only a small amount of cystic fluid remained (arrowhead). Intestinal loops were observed in the space which had been previously occupied by the huge cyst. A small amount of ascites was found in contact with the anterior surface of the liver (arrow). (A) Plain images, (B) Postcontrast images.

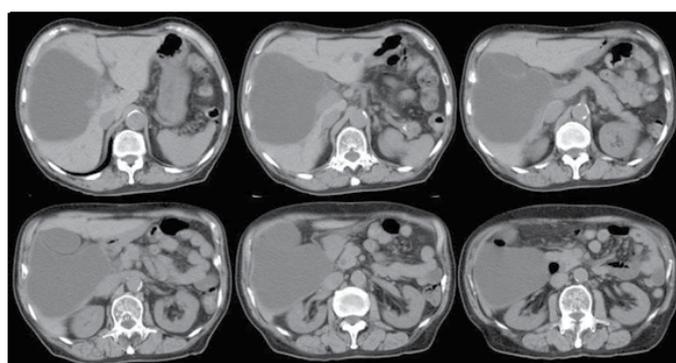


Figure 5: On July 15, 2009 the patient complained of back pain, and another computed tomography scan showing reappearance of the cyst, which grew to 13×10 cm in size.

[5, 6]. However, since our patient had no previous history of trauma or other episodes leading to mechanical injury to the cyst, the rupture was spontaneous [2, 7, 8]. This complication of hepatic cysts is a rare occurrence [1, 2].

In one report, spontaneous rupture was attributed to necrosis of the secretory cells by increased intracystic pressure, or ischemic necrosis due to pericystic scar

formation caused by local inflammation [7]. Some researchers observed intracystic hemorrhage prior to the rupture and speculated that the injured epithelial lining cells resulted in rupture [8]. In our case, there was no sonographic sign of intracystic hemorrhage on June 10, 2009, and the CT density of the remaining cystic fluid and ascites was not suggestive of hemorrhage. However, compared with the MR images in 2001 and the CT images in 2008, the cyst definitely increased in size and possibly became vulnerable to faint mechanical injury or insufficient blood supply to the lining cells which presumably primed the cyst for rupture.

There are a small number of reports on the sequential changes in the size of hepatic cysts during the obliteration process. In the case of cyst regression possibly through the disruption of the blood supply to the lining cells without rupture, a reduction in cyst diameter from 7.7 cm to 1.0 cm was observed during eight years of follow-up. Thus, the regression process seems to require a long-time [9]. However, cyst obliteration through rupture is reported to be sudden-onset and to rapidly progress. In our case, we speculated that the accumulation of a large amount of ascites just after rupture, its absorption by the peritoneal membrane and its clearance occurred before cyst disappearance [6]. This case suggests that the entire process can be completed only in nine days or less.

There are a few reports on the clinical course after the disappearance of hepatic cyst following rupture. In our case, cystic fluid accumulated again in less than one month, which suggested that the ruptured point was small and underwent spontaneous healing and closure.

Since re-rupture might cause bleeding inside the cyst or into the peritoneal cavity, prophylactic treatment was considered. Treatment options generally include radiological intervention by needle aspiration combined with injection of sclerosing agents or surgical approaches such as internal drainage with cystojejunostomy, wide deroofting and liver resection. Recent trends in the management of symptomatic hepatic cysts have shifted to minimally invasive procedures. Radiological intervention is safe, relatively noninvasive and considered as a first-line treatment especially for patients with high surgical risk or polycystic liver disease [1].

As a sclerosing agent, ethanol has been conventionally used, but it can cause pain, fever and intoxication. Some authors mentioned that the ideal quantity of ethanol is 25% of the total cystic aspirate and recommended that not greater than 100 mL of ethanol be injected at one time [4]. Although there was no leakage of intracystically injected contrast medium prior to ethanol injection, absolute ethanol may cause re-rupture of the repaired cystic wall during injection. Therefore, in the first session, only 70 mL of ethanol was injected. Since no problems arose following the first session, 100 mL of ethanol was injected in the second session. To compensate for the volume reduction of ethanol and to bring about the cooperative effect, we also used minocycline hydrochloride expecting its strong acidity to degenerate or kill secretory cells of

hepatic cyst [4, 10].

In our case, from April 30, 2008 to June 10, 2009, the patient did not complain of pain or discomfort probably because cyst enlargement gradually progressed. On the other hand, on July 15, 2009, although the volume of cystic fluid was less than that on April 30, 2008, he complained of back pain possibly because the regrowth process after rupture was rapid. Thus, if the hepatic cyst is not accompanied by bacterial or parasitic infection or intracystic hemorrhage, rupture can be asymptomatic, as seen in our case.

CONCLUSION

Simple hepatic cysts are generally stable in size over time. Spontaneous rupture of a simple hepatic cyst is a rare occurrence and the clinical course after rupture is not fully known. We radiologically demonstrated that the huge hepatic cyst was rapidly obliterated by spontaneous rupture and fast regrew during a one-month period. To our knowledge, this is the first case report which demonstrated such a rapid change in the morphology of the hepatic cyst during a short period after spontaneous rupture. In our case the patient was asymptomatic before and just after rupture and he complained of back pain only after the cyst regrew rapidly. Therefore, in some cases of huge hepatic cysts, transient episodes of rupture may be dismissed because it can be asymptomatic and the cystic fluid may have possibly already accumulated by the time symptoms of abdominal or back pain appear.

Author Contributions

Tomoki Nakajima – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Manabu Okajima – Conception and design, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Akiko Shibuya – Conception and design, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Junko Yamaoka – Conception and design, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Toshiaki Nakashima – Conception and design, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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