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Case report

Transcatheter arterial embolization for hemorrhagic rupture of a simple hepatic cyst: A case report[☆]

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ARTICLE INFO

Article history:

Received 15 April 2021

Revised 26 April 2021

Accepted 27 April 2021

Keywords:

Hepatic cyst hemorrhage

Hemorrhagic rupture of hepatic cyst

Symptomatic hepatic cyst

Transcatheter arterial embolization

Interventional radiology

ABSTRACT

Hemorrhagic rupture is a very rare and life-threatening hepatic cyst complication. Several treatment methods have been used for hepatic cyst hemorrhage and/or rupture; however, transcatheter arterial embolization for hepatic cyst hemorrhage has been poorly documented. An 80-year-old man receiving dual antiplatelet therapy was diagnosed with hemorrhagic rupture of a hepatic cyst. Transcatheter arterial embolization using a coil was performed for A6 branch confirmed active extravasation. His condition improved promptly after treatment, and the hepatic cyst gradually became smaller as compared to the size before hemorrhage. Transcatheter arterial embolization is suitable for hepatic cyst hemorrhage and might be a minimally invasive treatment option for a symptomatic hepatic cyst.

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Introduction

A hepatic cyst is typically asymptomatic liver tumor; however, it can sometimes cause various complications such as intracystic hemorrhage (2%–5% of cases), infection (1% of cases), and rupture (very rare) [1]. Hemorrhagic rupture is a very rare and life-threatening hepatic cyst complication [2]. Currently, no definitive etiology has been confirmed for hepatic cyst hemorrhage [3]. Hepatic cyst rupture is generally associated with a large volume increase [1]. Several treatment methods have been used for hepatic cyst hemorrhage and/or rupture;

however, there is no consensus on the optimal management strategy for a hepatic cyst hemorrhage [3]. Additionally, interventional radiology (IVR) for hepatic cyst hemorrhage has been poorly documented.

Here, we performed IVR for hemorrhagic rupture of a hepatic cyst and successfully treated the condition with transcatheter arterial embolization (TAE). TAE for hemorrhagic rupture of a hepatic cyst is extremely rare; to the best of our knowledge, this is the first report describing the clinical course of a simple hepatic cyst after TAE. In this report, we present our clinical experience and new findings with a review of the literature.

[☆] Competing Interests: The authors have no conflict of interest to declare.

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<https://doi.org/10.1016/j.radcr.2021.04.066>

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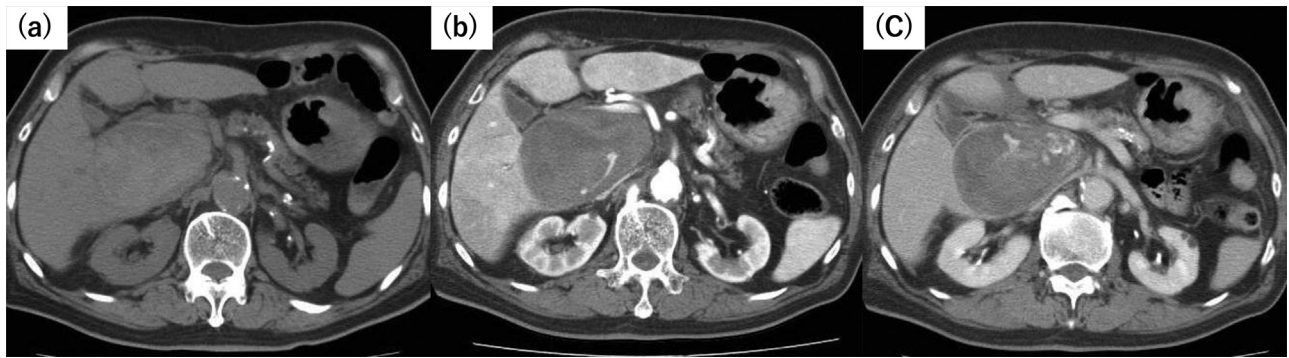


Fig. 1 – The CT findings at the emergency department.

- (A) Abdominal CT showed an enlarged, heterogeneous hepatic cyst (88 × 63 mm in size).
 (B) Extravasation in the hepatic cyst was shown on contrast-enhanced CT in early phase.
 (C) Extravasation was found throughout the cyst on contrast-enhanced CT in late phase.

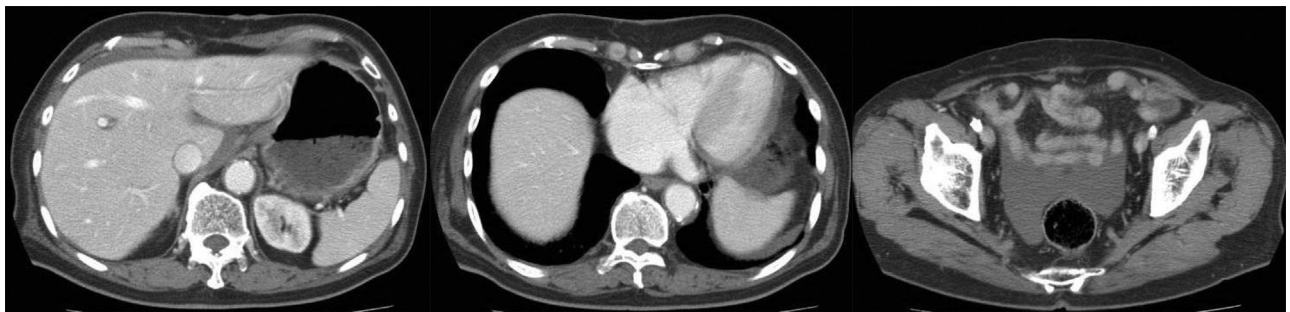


Fig. 2 – Contrast-enhanced CT revealed ascites in the entire abdominal cavity.

Case report

An 80-year-old man was referred to the emergency department because of shivering and fever. His surgical history included coronary artery bypass grafting, aortic valve replacement, and femoral-popliteal artery bypass. He received dual antiplatelet therapy (DAPT) with aspirin and clopidogrel. Additionally, he received regular rehabilitation therapy for heart disease in our hospital's outpatient department.

On laboratory assessment, his hemoglobin level was low (7.5 g/dL). Abdominal computed tomography (CT) showed an enlarged, heterogeneous hepatic cyst (Fig. 1A). Two hours later, contrast-enhanced CT revealed active extravasation in the hepatic cyst (Fig. 1B and C). Ascites was found throughout the peritoneal cavity on contrast-enhanced CT (Fig. 2); however, it was not found on the initial abdominal CT. Based on these findings, he was diagnosed with hemorrhagic rupture of hepatic cyst with active bleeding.

The patient was hemodynamically stable; therefore, we decided to perform IVR. The patient agreed with our decision to treatment. Under local anesthesia, the right femoral artery was cannulated using a 4-Fr long-sheath catheter. Hepatic arteriography was performed using a 4-Fr RH catheter, and selective angiography of the A6 branch confirmed active extravasation. TAE was performed using a coil (Azur 2 mm/2 cm, TERUMO). The findings of IVR are shown in Fig. 3. After TAE,

the arterial flow in the A6 branch disappeared. The patient received 4 units of erythrocytes after IVR.

The laboratory data showed an increased hemoglobin level 1 day after surgery, which indicated hemostasis. On contrast-enhanced CT, performed 5 days after surgery, extravasation in the hepatic cyst was not noted. On ultrasonography (US), the hemorrhagic hepatic cyst was recognized as a cyst with a distinct border and heterogeneous contents, similar to the CT findings. The patient's postoperative course was uneventful, and he was discharged 7 days after TAE.

In the outpatient department, we assessed the CT, and US findings for a hepatic cyst after TAE. On contrast-enhanced CT, the hepatic cyst gradually became smaller and homogeneous. Additionally, the hepatic cyst shrank 1 year after TAE when compared to the size before hemorrhage (Fig. 4). The patient is being followed through our hospital's outpatient department.

Discussion

Hepatic cysts can be divided into the following 2 general categories: Congenital and acquired [4]. Congenital hepatic cysts include simple hepatic cysts and polycystic liver disease (PCLD) [5]. Regarding simple hepatic cysts hemorrhage etiology, it has been hypothesized that, under high intracystic

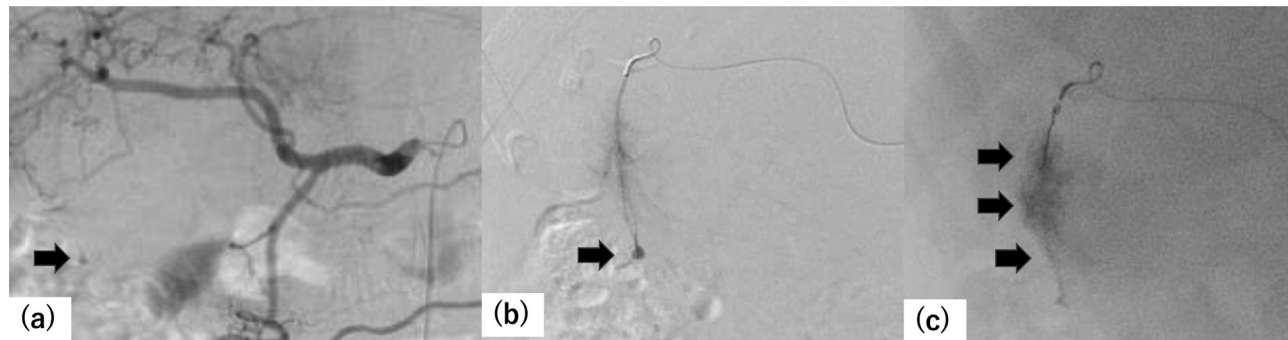


Fig. 3 – The findings of IVR.

(A) Angiography from common hepatic artery showed slight extravasation (black arrow).

(B) Selective angiography of the A6 branch confirmed active bleeding (black arrow).

(C) Contrast agent spread in the hepatic cyst (black arrow). TAE was performed using coil.

pressure, the cyst's epithelial lining undergoes necrosis and sloughing, injuring fragile blood vessels in the cystic wall and leading to intracystic hemorrhage [3,6,7]. Intracystic bleeding increases the tension inside the cyst and causes rupture with shock [8].

For a symptomatic simple hepatic cyst, conservative management, percutaneous drainage, sclerotherapy, surgical fenestration, and liver resection are common treatment options. More recently, minimally invasive therapeutic approaches have played increasing roles in managing cystic diseases of the liver [9]. In most centers, sclerotherapy is attempted first as a noninvasive option, and laparoscopic fenestration is usually indicated in refractory cases [10]. Currently, TAE is not performed for a symptomatic simple hepatic cyst. This is the first report of successful TAE for simple hepatic cyst hemorrhage rupture.

According to a review article on managing a bleeding liver tumor, when active bleeding is visualized, TAE is recommended over surgery unless there is severe hemodynamic instability [11]. In trauma practice guidelines, TAE is recommended for hemodynamically stable patients with evidence of active extravasation on imaging [12]. From these findings in other fields, the management strategy in our case is considered optimal. Our patient received DAPT, leading to a high risk of surgical complications such as intraoperative bleeding. Thus, TAE appears to have advantages over emergency surgery.

Hemorrhagic rupture of a hepatic cyst is a fatal condition. Imaoka et al. reported that 6 of 7 hepatic cyst rupture cases with intracystic bleeding required emergency surgery [8]. Marion et al. reported that 6 patients with hemorrhagic rupture of a hepatic cyst presented with hemodynamic decompensation, causing death in 3 patients [1]. In this case, hemorrhagic rupture of a hepatic cyst was successfully treated by TAE without hemodynamic failure. On the other hand, if there is no active extravasation on angiography, it may be difficult to select the treatment method. Ishikawa et al. reported on hepatic cyst hemorrhage with no active extravasation at the time of angiography [13]. They performed TAE for the peripheral vessels of a huge hepatic cyst suspected to be associated with bleeding; however, surgery was required later. The applicable

circumstances may be limited, and TAE for hepatic cyst hemorrhage requires more cases and further discussion.

The incidence rate of a hepatic cyst increases with age. Tenja reported that the overall prevalence rate was 5.81% and that the rates were 20.30% at the age of 50 years, 28.39% at the age of 60 years, and 38.46% at the age of more than 70 years [14]. It has been reported that anticoagulant or antiplatelet therapy does not appear to increase the risk of bleeding from liver tumors [11]. In contrast, there are some cases of hemorrhagic rupture of hepatic cysts with anticoagulation therapy, including our case [2,15,16]. Hepatic cyst hemorrhage may become severe in patients who receive anticoagulant or antiplatelet therapy. Given the future aging society, the number of elderly people with hepatic cysts and antithrombotic agent therapy is expected to increase [3,17], which may lead to more occurrences of hepatic cyst hemorrhage.

The distinction between hemorrhagic simple hepatic cysts and hepatobiliary cystic neoplasms can be difficult because both lesions have intracystic structures [5,6,13]. In hepatic cyst hemorrhage, US clearly shows intracystic blood clots as papillary, nodular tumorous or irregular septal images, whereas CT cannot demonstrate intracystic blood clots [18]. Discordance between US and CT findings has been proposed as an important element for diagnosing hepatic cyst hemorrhage vs hepatobiliary cystic neoplasm [3,18]. In the present case, identifying active extravasation on contrast-enhanced CT resulted in the diagnosis of hepatic cyst hemorrhage. On contrast-enhanced CT performed 5 days after TAE, the contrast effect of the hepatic cyst was not recognized. According to the experience of this case, if improvement is not obtained promptly, the image diagnosis must be reevaluated for the possibility of a cystic tumor. Magnetic resonance imaging (MRI) may help differentiate intracystic hemorrhage from other cystic lesions [19,20], because MRI images clearly show the morphologic features of blood clots in the cyst [18]. Repeated observation with CT or US is recommended as the most reliable method for distinguishing intracystic hemorrhage from a cystic neoplasm [19].

Our patient underwent repeated observation with CT and US as an outpatient. During outpatient follow-up after TAE, the hepatic cyst gradually reduced in size. To our knowledge,

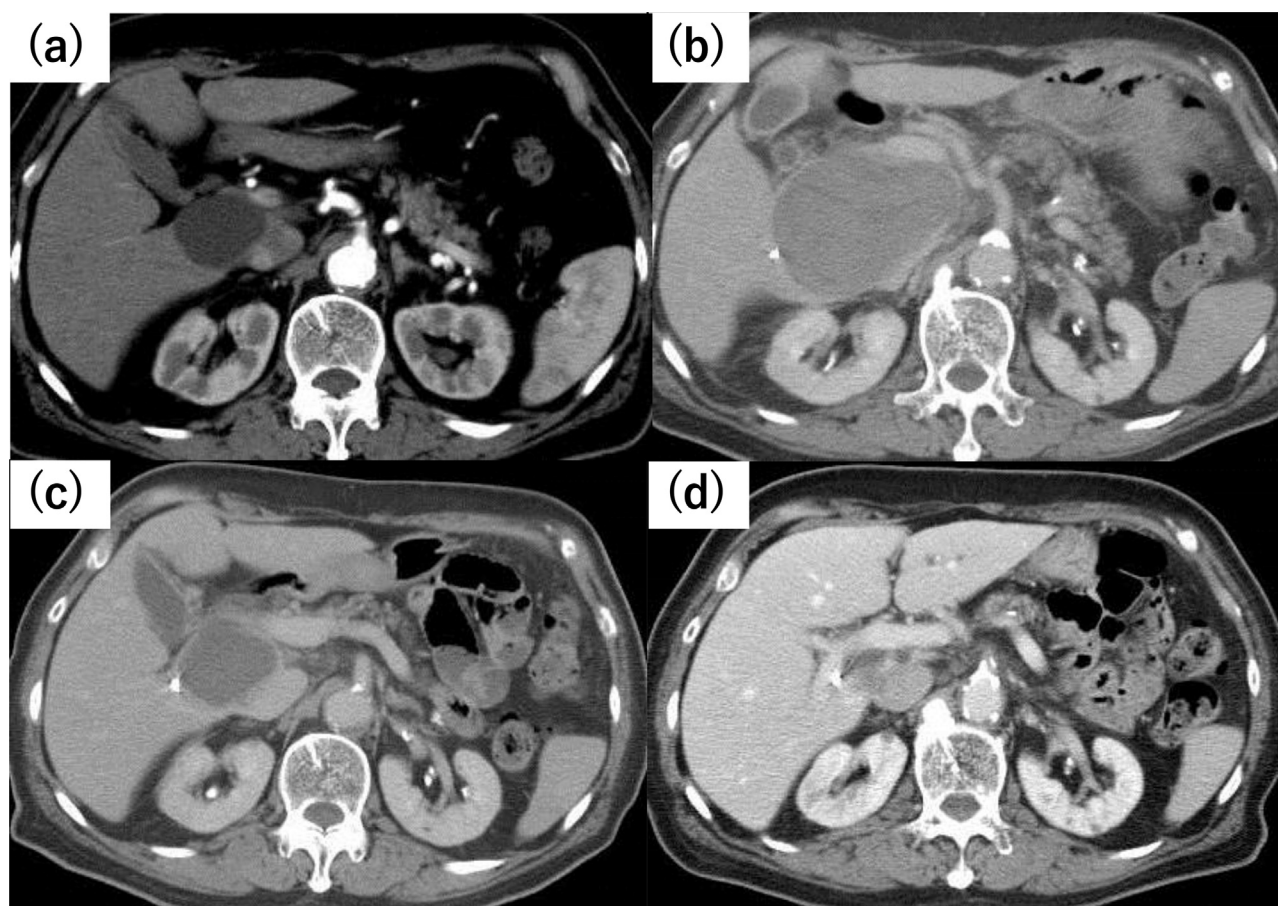


Fig. 4 – The clinical course of the hepatic cyst on contrast-enhanced CT findings.

(A) A simple hepatic cyst (43 × 35 mm in size) was found 1.5 years before hemorrhage.

(B) No active bleeding was shown 5 day's after TAE. The cyst (84 × 59 mm in size) was heterogeneous.

(C) The hepatic cyst (50 × 43 mm in size) became homogeneous 3 months' after TAE.

(D) The hepatic cyst shrank 1 year after TAE when compared to the size before hemorrhage (27 × 21 mm in size).

no report has described the course of a simple hepatic cyst following TAE. In limited facilities, TAE has become an accepted treatment option for patients with symptomatic PCLD [21]. In cystic lesions, almost all hepatic arterial branches are well developed, and they predominate over portal vein branches [22]. Therefore, TAE of hepatic artery branches that supply major hepatic cysts causes the cysts to shrink [23]. We hypothesized that a simple hepatic cyst is also artery dominant and that TAE shrinks the cyst by the same mechanism as in PCLD cases.

Basil et al. reported that radiologic drainage is reserved for a hepatic cyst in the posterior segment, where access with laparoscopy may be limited [9]. On the other hand, TAE is not restricted in any segment. Therefore, TAE might be an option for cases that surgical approach is difficult.

In summary, hemorrhagic rupture of a simple hepatic cyst was treated successfully with TAE. The hepatic cyst size reduced after TAE. In a future aging society, the incidence of hepatic cyst hemorrhage may increase, and various treatment options will be required. TAE might be a versatile and minimally invasive treatment option for hemorrhagic rupture of a hepatic cyst. Further discussion of appropriate TAE for a symptomatic simple hepatic cyst is necessary while referring to PCLD cases.

Patient consent

The written informed consent was obtained for publication.

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