

# Delayed Intracystic Hemorrhage after Percutaneous Drainage and Sclerotherapy for a Symptomatic Giant Hepatic Cyst: A Case Report

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## Abstract

Herein, we have reported a rare case of intracystic hemorrhage due to rupture of a right hepatic artery pseudoaneurysm in a 76-year-old female patient who underwent drainage and 3% polidocanol sclerotherapy for a symptomatic giant hepatic cyst. One month after sclerotherapy, the patient presented to the emergency room with acute and severe abdominal pain. Non-contrast T1-weighted magnetic resonance imaging findings showed high hepatic cyst fluid signal intensity and abdominal arteriography findings revealed a right hepatic artery pseudoaneurysm surrounding the hepatic cystic wall. Therefore, the patient was diagnosed with intracystic hemorrhage due to a ruptured pseudoaneurysm. Embolization, using a detachable coil, was successful. Interventional radiologists should be aware of potential vascular injuries during drainage and sclerotherapy for giant hepatic cysts.

**Key words:** symptomatic hepatic cyst, sclerotherapy intracystic hemorrhage, pseudoaneurysm, embolization

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## Introduction

Percutaneous drainage followed by sclerotherapy is the currently preferred noninvasive treatment approach for sustained volume reduction and symptomatic relief in patients with hepatic cysts [1]. Although this treatment approach is associated with few serious complications, several studies have recently reported intracystic bleeding during and after the procedure [2, 3]. Herein, we have reported a rare case of intracystic hemorrhage due to rupture of a right hepatic artery pseudoaneurysm after drainage and 3% polidocanol sclerotherapy in a patient with a symptomatic giant hepatic cyst.

## Case Report

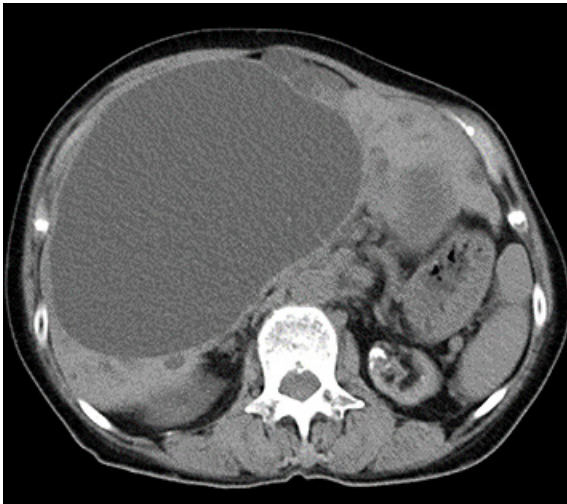
A 76-year-old woman presented with epigastric discom-

fort and abdominal distention after eating. Non-contrast computed tomography (CT) revealed compression of the descending duodenum by a giant hepatic cyst measuring  $17 \times 12 \times 17$  cm in segment 4 of the liver (**Fig. 1**). Percutaneous drainage and ethanol sclerotherapy are commonly used [4]; however, polidocanol sclerotherapy and ethanol sclerotherapy have been recently reported to have similar therapeutic effects, although ethanol sclerotherapy has more side effects than polidocanol sclerotherapy [2]. Therefore, percutaneous drainage and 3% polidocanol sclerotherapy were performed to relieve the symptoms. Briefly, using the right hypochondriac approach, the hepatic cyst was punctured through segment 5 of the liver with an 18-gauge puncture needle during ultrasonography. A 7-Fr pigtail catheter was placed and a 0.035-inch spring guide wire inserted into the cyst under fluoroscopic guidance, and 1800 mL of serous fluid was collected through the drainage catheter (**Figs. 2 and 3**). Subsequently, sclerotherapy was performed using 6

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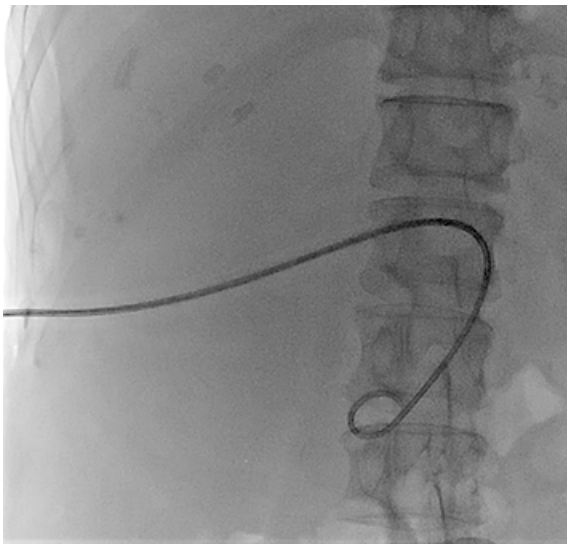
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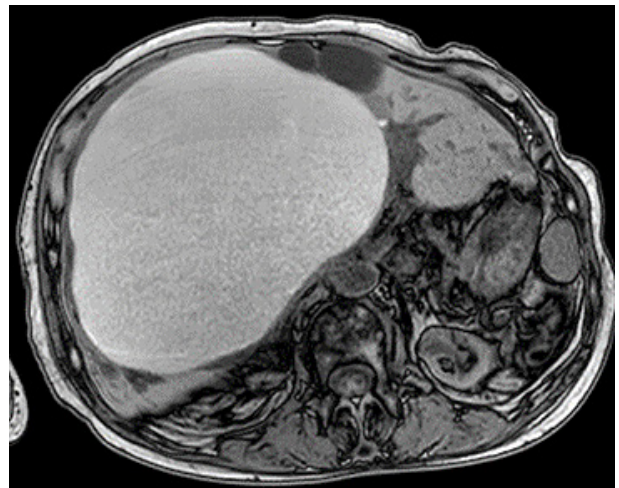
**Figure 1.** Non-contrast CT shows a large hepatic cyst, 17 × 12 × 17 cm in dimensions, in segment four of the liver.



**Figure 3.** Non-contrast CT after fluid drainage of the hepatic cyst shows the pig-tail drainage catheter (arrow) positioned in the liver hilum.



**Figure 2.** X-ray fluoroscopy shows a 7F drainage catheter inserted into the hepatic cyst through segment five of the liver using right hypochondriac approach.

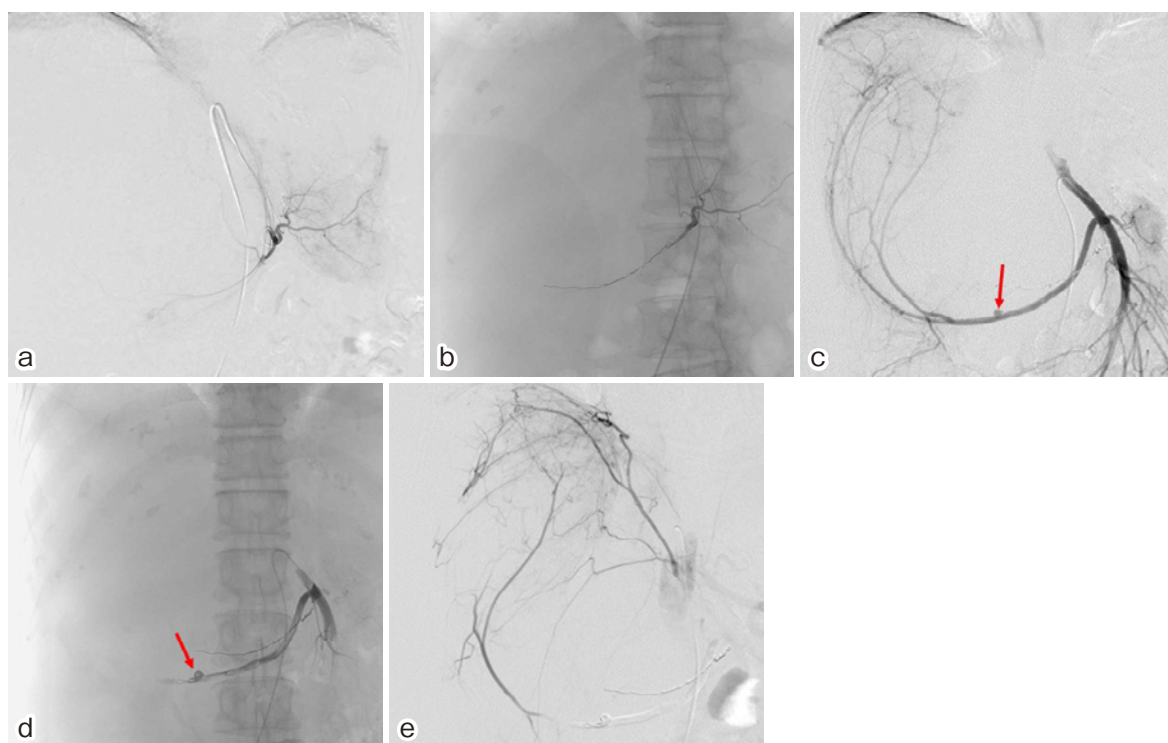


**Figure 4.** T1-weighted magnetic resonance image during sclerotherapy shows homogeneous high-intensity fluid of the hepatic cyst.

mL of 3% polidocanol foam with 24 mL of room air. No immediate issues were noted, and the catheter was kept in place for 24 h and removed thereafter. No hemorrhagic fluid was observed during the removal of the drainage catheter, and the patient was discharged five days later.

One month after sclerotherapy, non-contrast CT showed a 34% reduction in the volume of the hepatic cyst compared with its volume measured on pretreatment non-contrast CT images, and the patient reported symptomatic relief. Laboratory test results on the day were not abnormal, with a hemoglobin level of 9.9 g/dL, leukocyte count of 4800 cells/ $\mu$ L, and C-reactive protein (CRP) level of 1.15 mg/dL. However, the patient presented to our emergency room with a seven-day history of fever (body temperature: 38°C) and sudden onset of abdominal pain in the right hypochondrium. On arrival, her blood pressure was 112/57 mmHg, and her heart

rate was 57 beats per minute. The laboratory test results on admission were as follows: hemoglobin level, 8.6 g/dL; leukocyte count, 4900 cells/ $\mu$ L; platelet count,  $17.6 \times 10^4$  cells/ $\mu$ L; estimated glomerular filtration rate (eGFR), 10.3 mL/min; and CRP level, 9.69 mg/dL. The hemoglobin level further dropped to 7.1 g/dL after two days. Non-contrast CT revealed enlargement of the treated hepatic cyst compared with its size measured on images obtained seven days earlier. Contrast-enhanced CT could not be performed because of poor renal function, and T1- and T2-weighted non-contrast magnetic resonance imaging revealed high signal intensity of the treated hepatic cyst (**Fig. 4**). Intracystic hemorrhage and infection were suspected. The patient had persistent right hypochondrial pain and worsening anemia. The cause of the intracystic hemorrhage was unknown; therefore, abdominal angiography was performed for transcatheter arterial embolization (TAE) of the hepatic arteries feeding the



**Figure 5.** a. Left hepatic arteriography shows no extravasation from the middle hepatic artery. b. Left hepatic arteriography shows embolization of the middle hepatic artery using pushable-coils. c. Superior mesenteric arteriography shows a pseudoaneurysm (arrow) in the replaced right hepatic artery originated from the superior mesenteric artery. d. Superior mesenteric arteriography after coiling of the pseudoaneurysm shows embolization of the pseudoaneurysm using a detachable coil (arrow) (Target XL; diameter, 6 mm; length, 20 cm; Stryker Corporation, Tokyo, Japan). e. Right infraphrenic arteriography shows the presence of sufficient collateral arteries to feed the right hepatic lobe via the right infraphrenic artery.

cystic wall under sufficient hydration.

Left hepatic arteriography showed no extravasation from the middle hepatic arteries (**Fig. 5a**); however, the middle hepatic arteries were embolized to reduce the cyst size (**Fig. 5b**). Superior mesenteric artery arteriography performed thereafter revealed a pseudoaneurysm in the right hepatic artery (**Fig. 5c**), and coil embolization using a detachable coil (Target XL; diameter, 6 mm; length, 20 cm; Stryker Corporation, Tokyo, Japan) was immediately performed (**Fig. 5d**). Right infraphrenic arteriography revealed the presence of sufficient collateral arteries to feed the right hepatic lobe via the right infraphrenic artery (**Fig. 5e**). The right hypochondrial pain and abdominal distention disappeared after TAE, and no progression of anemia was observed. The patient was discharged 14 days after TAE, and six months later, abdominal ultrasound revealed a marked reduction in the cyst size, which was  $9 \times 6$  cm in diameter.

## Discussion

Most congenital hepatic cysts, including simple cysts and polycystic liver disease, which have an approximate prevalence of 18% in adults, are asymptomatic [1]. Enlargement of hepatic cysts during follow-up may be associated with

symptoms and complications such as pain, nausea, meteorism, vomiting, early satiety, intracystic infection, and obstructive jaundice [1]. The long-term efficacy and safety of percutaneous drainage and sclerotherapy for symptomatic hepatic cysts have been demonstrated [4]. However, intracystic hemorrhage may occasionally occur as a life-threatening complication in 2%-5% patients [5]. In a review of case reports on hemorrhagic hepatic cysts published in 2015, 15 of 28 (54%) patients underwent surgical treatment, including partial hepatectomy (n = 8, 29%), cystectomy (n = 3, 11%), and fenestration (n = 4, 14%), whereas seven (25%), two (7%), and four (14%) patients underwent transhepatic percutaneous drainage, TAE, and conservative therapy, respectively [5-9]. The etiology and incidence of intracystic hemorrhage are unclear. The hepatic cystic wall includes three layers—an inner single layer comprising cuboidal or columnar epithelium such as biliary epithelial cells, a middle layer comprising compact connective tissue containing small blood vessels, and an outer layer comprising loose connective tissue with large blood vessels, bile ducts, and occasional von Meyenburg complexes. Increased intracystic pressure due to an increase in cyst volume because of biliary epithelial secretion causes necrosis and sloughing of the biliary epithelium, leading to the exposure of blood vessels



within the middle or outer cystic wall layer. Subsequent injury to the blood vessels in the fragile cystic wall may be responsible for intracystic hemorrhage. Furthermore, rupture of the fragile cystic wall due to increased intracystic pressure because of rapid intracystic hemorrhage can lead to serious outcomes.

TAE can produce therapeutic effects in patients with poor clinical conditions who cannot tolerate surgical treatment; however, bleeding may recur afterwards. Ishikawa *et al.* reported a case of a patient who underwent TAE for hepatic intracystic hemorrhage; however, the volume of the cyst increased three weeks after TAE, requiring simple cystectomy [8]. Identifying the source of bleeding within the hepatic cystic wall is often difficult using arteriography, and treatment with TAE may be unsuccessful. However, Takei *et al.* reported that TAE may be considered in patients with autosomal dominant polycystic kidney disease and symptomatic polycystic liver who are not candidates for surgical treatment because blood flow in the hepatic cystic wall is derived primarily from the hepatic artery [10]. Therefore, we suggest that TAE might be an effective treatment for symptomatic hepatic cysts if all intrahepatic arteries feeding the hepatic cystic wall are embolized; however, the efficacy of TAE for hemorrhagic hepatic cysts should be investigated in future studies.

To date, only two cases of iatrogenic intracystic hemorrhage during percutaneous drainage and sclerotherapy have been reported [2, 3]. Spârchez *et al.* reported a case of patient who experienced intracystic bleeding with clot formation during aspiration [2]. As sclerotherapy is contraindicated in patients with active bleeding, sclerotherapy was postponed for one month, and the patient was successfully treated. Percutaneous aspiration using an 18-G aspiration needle may lead to bleeding if the tip of the needle used during the procedure sticks to the cystic wall. Therefore, small catheters measuring 6-7 Fr are preferred over aspiration needles. Additionally, Sekiguchi *et al.* reported that complications such as penetration of the hepatic artery and portal vein occurred during sclerotherapy in a patient with symptomatic hepatic cysts [3]. They embolized the left hepatic artery from the arterial side with a micro-coil, injected 1.1 mL of 50% n-butyl-cyanoacrylate into the hepatic cyst from a 4-Fr straight catheter through the drainage catheter, and sealed the portal vein hole from the outside with a glue ball while removing the catheter system. In our case, the cause of the pseudoaneurysm in the replaced right hepatic artery was unknown because intracystic hemorrhage was not observed during the procedure, and the bleeding point was different from the puncture line. To date, no study has reported the formation of hepatic artery pseudoaneurysms around the cystic wall. We assumed that contact stimulation

of the drainage catheter during overnight placement damaged the exposed hepatic artery surrounding the hepatic cystic wall and caused pseudoaneurysm in the right hepatic artery. In our case, the pseudoaneurysm was safely embolized using an isolation technique.

In conclusion, interventional radiologists should be aware of the potential damage to the hepatic arteries in the middle or outer layer of the cystic wall during needle puncture or insertion of a drainage catheter. Urgent embolization may be needed in patients with artery injuries. TAE of the damaged hepatic arteries feeding the hepatic cystic wall is an effective method for the treatment of intracystic hemorrhage.

**Conflict of interest:** The authors declare that they have no conflicts of interest to report.

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