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Intracystic Hemorrhage Complicating Polycystic Liver Disease in a 90-Year-Old Woman

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ABSTRACT

Nonparasitic hepatic cysts are common benign tumors that are often asymptomatic and incidentally discovered on imaging. Intracystic hemorrhage is a rare complication of hepatic cysts. We review the literature and discuss a case of intracystic hemorrhage in a 90-year-old woman with polycystic liver disease. The patient underwent cyst aspiration and percutaneous drain placement with subsequent resolution of symptoms. To our knowledge, we report the oldest patient to present with hemorrhage into a hepatic cyst. This case presents unique challenges in management, both because of the patient’s age and because of her polycystic liver disease.

INTRODUCTION

Simple hepatic cysts are benign lesions that are often asymptomatic and discovered incidentally on imaging, with a prevalence of 5%–15%.1,2 Mass effect and compression of adjacent structures are the most frequent complications, and most patients present with abdominal pain.3 In rare cases, they can be complicated by intracystic hemorrhage. After an extensive literature search, we identified 23 publications describing 27 cases of nonruptured hemorrhagic hepatic cysts and only 3 cases in patients with polycystic liver disease (PCLD). The mean age at presentation was 60 years. We report a case of intracystic hemorrhage in a 90-year-old woman with isolated PCLD. This is the oldest patient reported to present with a hemorrhagic hepatic cyst.

CASE REPORT

A 90-year-old woman with chronic thrombocytopenia, heart failure, and atrial fibrillation on warfarin presented with acute-on-chronic abdominal pain. She had multiple hepatic cysts, which were incidentally discovered 9 months previously. The largest cyst was 11 cm. She presented with mild abdominal discomfort that had acutely progressed to sharp right upper quadrant abdominal pain. Two months previously, she underwent transcatheter aortic valve replacement for aortic stenosis and was started on clopidogrel.

The patient was hemodynamically stable and had a large, palpable mass in the right upper quadrant with severe localized tenderness to palpation. Liver function tests, hemoglobin, and leukocytes were within normal limits, platelets were 42,000/mL, and international normalized ratio was 1.5. Computed tomography angiography revealed a polycystic liver with a dominant 11-cm exophytic cyst containing dependently layering hyperdense material consistent with intracystic hemorrhage (Figure 1). There was no active extravasation.

The patient’s warfarin was held, and she was given 1 unit of fresh-frozen plasma and platelets. Given her age and multiple medical comorbidities, she was deemed a poor surgical candidate. Because the patient continued to have severe abdominal pain refractory to
analgesics, the decision was made to proceed with drainage. Interventional radiology placed a percutaneous drain under ultrasound and fluoroscopic guidance and aspirated 500 mL of blood from the cyst (Figure 2). The patient had subsequent resolution in symptoms and remained hemodynamically stable with stable hemoglobin. She then returned to interventional radiology for cyst sclerotherapy 2 days later. Contrast injection into the cyst under fluoroscopy demonstrated extravasation,

Figure 1. (A) Abdominal computed tomography angiography revealed an 11 × 9 cm exophytic cyst arising from the inferior right hepatic lobe containing dependently layering hyperdense material (arrow), consistent with intracystic hemorrhage. (B) Coronal and (C) sagittal views provide visualization of the patient’s polycystic liver.

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precluding sclerosis due to the concern of sclerosant extravasa-
tion (Figure 3). The drain was left in place for planned reat-
tempt at sclerotherapy in 1 month. In the interim, the patient’s
drain became dislodged. One month later, ultrasound showed
that the cyst had decreased to 7.8 × 7.0 cm and had minimal
residual hyperechoic material, consistent with drainage and
resorption of the previously seen hemorrhage (Figure 4). She
remains asymptomatic 11 months later.

DISCUSSION
Nonparasitic benign liver cysts are usually asymptomatic and
can be congenital or acquired, possibly arising from biliary

Common presenting symptoms of hemorrhagic hepatic cysts
include abdominal pain, palpable abdominal mass, and ob-
structive jaundice.9 Diffuse peritoneal signs, hemodynamic
instability, and progressive anemia are signs of hemorrhagic
cyst rupture, which is a rare, life-threatening complication that
occurs preferentially in patients with PCLD.10 Thirteen cases of
hemorrhagic rupture have been reported in the literature,
resulting in 4 deaths, whereas no deaths secondary to intracystic
hemorrhage without rupture have been reported.6,7,10 Preventing
rupture is therefore the top priority in the acute man-
agement of contained intracystic hemorrhage. Computed
tomography angiography should be performed to assess for
extravasation, which increases the risk of rupture and should be
treated with urgent angioembolization.

Minimizing recurrence has proven to be one of the major chal-
lenes in the management of hepatic cysts. Lo he et al observed
a 67% and 41% symptomatic recurrence rate in patients with
PCLD and simple liver cysts after surgical unroofing, resection,
or transplant.11 Surgical unroofing is the definitive choice for
single symptomatic hepatic cysts because it effectively relieves
symptoms, has a low recurrence rate, and allows for analysis of
the cyst wall.9,12,13 However, percutaneous drainage followed by
injection of sclerosing agents is the preferred option, particularly
for poor surgical candidates or those who prefer a less invasive
approach. It has been reported that this is as effective as
Table 1. Gigot classification of patients with adult polycystic liver disease¹²⁻¹⁵

<table>
<thead>
<tr>
<th>Type</th>
<th>Definition</th>
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<tbody>
<tr>
<td>Type I</td>
<td>Limited number (&lt;10) of large cysts (&gt;10 cm)</td>
</tr>
<tr>
<td>Type II</td>
<td>Diffuse involvement of the liver parenchyma by multiple medium-sized cysts with remaining large areas of the noncystic liver parenchyma</td>
</tr>
<tr>
<td>Type III</td>
<td>Massive, diffuse involvement of the liver parenchyma by small- and medium-sized liver cysts, with only a few areas of the remaining normal liver parenchyma between cysts</td>
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</table>

Surgery is indicated when malignancy is suspected, in the presence of a communication between the cyst and biliary tree, and in cases of failed nonoperative management. When approaching these patients, it is helpful to use the Gigot classification as a guide (Table 1).¹²⁻¹⁵ In a subset of patients with Type I PCLD who are symptomatic from 1 dominant cyst, laparoscopic unroofing is the procedure of choice. In patients with Type II and III PCLD presenting with symptomatic hepatomegaly, hepatic resection and cyst fenestration in the remnant liver is a more appropriate option.

Our patient had resolution of symptoms after percutaneous drainage and remains asymptomatic 11 months later. This supports previous reports suggesting that percutaneous drainage and sclerotherapy is safe and effective and should be performed as the first-line treatment of symptomatic hepatic cysts, even in the presence of PCLD and/or acute intracystic hemorrhage.¹²⁻¹⁴ Patients with suspected malignancy, communication between the cyst and biliary tree, or recurrence of symptoms after percutaneous drainage and sclerotherapy should undergo surgery.

REFERENCES


DISCLOSURES

Author contributions: AC Purdy performed the literature review and wrote the manuscript. A. Grigorian, D. Fernando, J. Nahmias, and AN Demirjian edited the manuscript. AN Demirjian is the article guarantor.

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Informed consent was obtained for this case report.

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