CASE REPORT

Nonparasitic Solitary Giant Hepatic Cyst Causing Obstructive Jaundice was Successfully Treated with Monoethanolamine Oleate

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Abstract

A 77-year-old man hospitalized for epigastric pain showed jaundice of the skin and conjunctivae. Laboratory tests revealed elevated hepatobiliary enzymes and inflammatory markers, and imaging studies demonstrated a 12 cm hepatic cyst compressing the common bile duct. The diagnosis was a giant hepatic cyst causing obstructive jaundice. Cyst drainage and sclerotherapy with 5% monoethanolamine oleate was performed twice, resulting in almost complete disappearance of the cyst. Obstructive jaundice due to a hepatic cyst, as seen in this case, is relatively rare and this report includes a review of other similar cases in Japan.

Key words: hepatic cyst, obstructive jaundice, monoethanolamine oleate

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Introduction

Hepatic cysts are usually asymptomatic but may occasionally present as abdominal pain, nausea, vomiting, and abdominal distention (1, 2). However, even in symptomatic hepatic cysts, obstructive jaundice is rarely seen. Sanfelippo et al (3) reported obstructive jaundice in only two of 82 patients with hepatic cysts. Recent trends in the treatment of symptomatic hepatic cysts, except in cases of acute rupture, hemorrhage or where cancer is suspected, include cyst drainage followed by drug injection (sclerotherapy). Ethanol and minocycline are often used as sclerosing solutions, and more recently, monoethanolamine oleate has been used with good results (4-6). We recently encountered a patient with obstructive jaundice due to a hepatic cyst who was successfully treated with sclerotherapy using monoethanolamine oleate. This case is presented here, together with a discussion of the related medical literature.

Case Presentation

A 77-year-old man was referred to our hospital because of persistent epigastric pain. His past history was unremarkable except for appendectomy at age 18 and pulmonary tuberculosis at age 75. Physical examination on admission showed the patient to be lucid and afebrile. His blood pressure was 152/84 mmHg and pulse rate 87 bpm. The abdomen was soft and slightly distended, with mild tenderness in the upper abdomen. There was no rebound, guarding, hepatosplenomegaly, palpable masses, or lower extremity edema. Laboratory findings on admission revealed elevated hepatobiliary enzymes and inflammatory markers (Table 1). Urinalysis was positive for bilirubin, and abdominal ultrasound showed a large cystic lesion in the right hepatic lobe (Fig. 1). The lesion contained no septum or calcifications, and the intrahepatic bile ducts in both hepatic lobes were dilated. Abdominal computed tomography confirmed the presence of a large cystic lesion 12 cm in diameter (Fig. 2a),

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which showed no enhancement with contrast medium (Fig. 2b). Both T1- and T2-weighted magnetic resonance images of the lesion showed homogeneously high signal intensity as compared with normal liver parenchyma (Fig. 3a and b). Magnetic resonance cholangiopancreatography indicated the presence of a large spherical lesion near the confluence of the right and left hepatic ducts that was compressing the intrahepatic bile ducts and keeping them separate from the common bile duct (Fig. 3c). Endoscopic retrograde pancreatography revealed downward compression of the common bile duct, gall bladder, and cystic duct (Fig. 4). The intrahepatic bile ducts could not be visualized on endoscopic retrograde pancreatography. No flow of contrast medium was detected in the cystic lesion, and neither gall stones, tumors, nor abnormalities of the pancreaticobiliary duct could be identified. On the basis of these findings, a diagnosis was made of obstructive jaundice due to a giant hepatic cyst.

On day 15 after admission, about 600 mL of fluid was drained from the cyst by means of transcutaneous transhepatic drainage. Repeat abdominal ultrasound showed almost complete disappearance of the cyst (Fig. 5). Sixty milliliters of 5% monoethanolamine oleate was then injected, the patient was placed in different positions for 30 minutes, after which the monoethanolamine oleate was aspirated. On the following day the drainage tube was removed. The fluid drained from the cyst was reddish-brown and serous, with cell counts of less than 100 (cells could not be classified), a specific gravity of 1.019, negative Rivalta test result, and cytologically identified as class I. Bacteriologic cultures of the cyst contents were negative, but cyst fluid tumor markers were markedly elevated: CEA, 94.7 ng/mL; CA19-9, ≥ 5,000 U/mL; CA125, 1,159 U/mL. Despite the initial almost complete disappearance of the cyst, regrowth was noted 28 days after the first drainage (Fig. 6), and a second drainage was thus performed. Drainage of about 300 mL of fluid followed by injection of 60 mL of monoethanolamine oleate resulted in disappearance of the hepatic cyst and resolution of the abdominal pain. On days 33 and 41 after admission, serum transaminase and total bilirubin, respectively, were within normal limits. On day 41 after admission, the patient was discharged from hospital. He has since shown an uneventful course with no recurrence of the hepatic cyst.

### Discussion

The prevalence of hepatic cysts is 0.1 to 0.5% (3) based on autopsy studies and 2.5% based on ultrasound examinations (7). Hepatic cysts have been classified by Henson et al (8) into four types: congenital, neoplastic, inflammatory, and traumatic. Congenital hepatic cysts are further categorized into solitary and polycystic cysts. The findings of the present patient are consistent with a solitary unilocular cyst.

For diagnosing a hepatic cyst, it is important to rule out hepatic cystadenoma or cystadenocarcinoma. Imaging study findings that suggest a cystadenoma or cystadenocarcinoma include the presence of solid elements with enhancement on contrast computed tomography (9). In the present patient, none of the imaging studies showed solid elements, and no enhancement was seen on contrast computed tomography. The diagnosis in this case was therefore a simple hepatic cyst. Laboratory findings on admission showed elevated hepatobiliary enzymes and inflammatory markers. In addition, serum transaminase was elevated, but normalized after cyst drainage. Since other causes of an elevated serum transaminase level were absent, this elevation was considered to be caused by obstructive jaundice.

Typical magnetic resonance imaging findings of a hepatic cyst include the same signal intensity as water (10), that is, homogeneous low signal intensity on T1-weighted imaging and homogeneous high signal intensity on T2-weighted imaging. However, our patient had homogeneous high signal intensity on both T1- and T2-weighted images, which suggests intracystic hemorrhage or a high protein concentration. This, combined with the reddish-brown color of the drained cyst fluid, indicates that the patient may have been suffering intracystic bleeding, which caused cyst enlargement prior to the development of symptoms, even though findings of abdominal ultrasound and abdominal computed tomography were not typical for intracystic hemorrhage.

Markedly elevated tumor marker levels have been reported even in histologically diagnosed simple cysts (11, 12). Iwase et al (13) reported CA19-9 levels in hepatic cyst fluid at least 100 times higher than in normal serum concentrations. The present patient also had markedly elevated cyst fluid tumor marker levels, and imaging studies indicated a simple cyst.

Solitary nonparasitic cysts of the liver causing obstructive jaundice were first reported in 1950 by Caravati et al (14). In connection with the findings for our patient, we reviewed reports of similar cases in the Japanese and English medical literature. We found a total of 51 patients with the following characteristics: mean age, 65.8±15.2 years (range, 29 to 90 years); male to female ratio, 7:6; cyst size, 12.5±5.1 cm (range, 2 to 30 cm); and total bilirubin, 10.2±7.7 mg/dL (range, 1.5 to 31.5 mg/dL). Twenty-nine patients were treated surgically and 22 were treated non-surgically. Of the

<table>
<thead>
<tr>
<th>Urine</th>
<th>Blood chemistry</th>
</tr>
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<tbody>
<tr>
<td>Protein (-)</td>
<td>AST 430 IU/L</td>
</tr>
<tr>
<td>Sugar (-)</td>
<td>ALT 492 IU/L</td>
</tr>
<tr>
<td>Occult blood (-)</td>
<td>7-GTP 692 IU/L</td>
</tr>
<tr>
<td>Bilirubin (3a)</td>
<td>LDH 340 IU/L</td>
</tr>
<tr>
<td>Peripheral blood</td>
<td>ALP 1884 IU/L</td>
</tr>
<tr>
<td>WBC 10600/mm³</td>
<td>T.Bil 7.0 mg/dl</td>
</tr>
<tr>
<td>Neutro 75.6%</td>
<td>D.Bil 6.0 mg/dl</td>
</tr>
<tr>
<td>Lym 17.1%</td>
<td>AMY 72 IU/L</td>
</tr>
<tr>
<td>Mo 62%</td>
<td>TP 6.5 g/dl</td>
</tr>
<tr>
<td>Ea 0.9%</td>
<td>APTT 26.1 sec.</td>
</tr>
<tr>
<td>Ba 0.2%</td>
<td>CEA 2.2 ng/ml</td>
</tr>
<tr>
<td>Hemoglobin 14.7 g/dl</td>
<td>CA19-9 10 U/ml</td>
</tr>
<tr>
<td>RBC 46810/mm³</td>
<td>Ch-E 274 I/U</td>
</tr>
<tr>
<td>Hb 44 %</td>
<td>CRP 2.8 mg/dl</td>
</tr>
<tr>
<td>Ht 25.2±10³/mm³</td>
<td>ESR 25 mm/hr</td>
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<tr>
<td>Ptt 14.7 g/dl</td>
<td>Hbs Ag (-)</td>
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Figure 1. Abdominal ultrasound. A large cystic lesion is located in the right hepatic lobe. The content is homogeneous, without a septum or calcifications, and the intrahepatic bile ducts are dilated.

Figure 2. Upper abdominal computed tomography. a: Plain computed tomography shows a large 12 cm cystic lesion. b: Contrast computed tomography shows no internal enhancement and no enhancement of the cyst wall.

patients for whom detailed medical records of surgery were available, 11 underwent cystectomy, 9 cyst fenestration, and 5 hepatic resection. Of those who were treated nonsurgically, 6 received only drainage and 16 received drainage followed by drug injection (sclerotherapy), using absolute ethanol and minocycline for 7 patients each, and monoethanolamine oleate for 2 patients.

The prognosis of solitary nonparasitic cysts of the liver with obstructive jaundice is relatively good. Irrespective of whether treatment is surgical or non-surgical, the patient should have a good clinical course. In the present case, we selected non-surgical treatment for the initial management in consideration of the patient’s quality of life.

Selection of the appropriate sclerotherapy type after cyst drainage is also important. Ethanol can cause abdominal pain (15), and in some cases overdosage may lead to alcoholic intoxication (16), while adverse reactions including eosinophilia have been reported for minocycline (17). Great care must thus be taken when using these drugs. Monoethanolamine oleate is commonly used for sclerotherapy of esophageal varices. Iwasaki et al (6) have treated at least 20 patients with symptomatic hepatic cysts using monoethanolamine oleate as “cyst sclerotherapy” and reported resolution or reduced size of the cyst in all cases without any adverse reactions or recurrence. As the cyst showed regrowth following initial treatment with monoethanolamine oleate, a second drainage was needed. This regrowth was thought to be caused by insufficiency of monoethanolamine oleate injection. However, thereafter, treatment with monoethanolamine oleate was effective for our patient. As monoethanolamine oleate sclerotherapy for hepatic cysts has been reported in only relatively few cases, further studies with a larger number of patients are necessary.

In the present case, the dose of injected monoethanola-
Abdominal magnetic resonance imaging. a: T1-weighted imaging shows a cystic lesion with homogeneous high signal intensity. b: T2-weighted imaging also shows homogeneous high signal intensity. c: Magnetic resonance cholangio-pancreatography indicates the presence of a large spherical lesion near the confluence of the right and left hepatic ducts that is compressing the intrahepatic bile ducts and keeping them separate from the common bile duct.

Figure 4. Endoscopic retrograde pancreatoc cholangiography. Downward compression of the common bile duct, gall bladder, and cystic duct. No contrast medium is seen in the intrahepatic bile ducts.

Figure 5. Upper abdominal computed tomography. The cystic lesion disappeared after drainage and monoethanolamine oleate treatment.

Magnetic resonance cholangio-pancreatography indicated the presence of a large spherical lesion near the confluence of the right and left hepatic ducts that is compressing the intrahepatic bile ducts and keeping them separate from the common bile duct.

Figure 3. Abdominal magnetic resonance imaging. a: T1-weighted imaging shows a cystic lesion with homogeneous high signal intensity. b: T2-weighted imaging also shows homogeneous high signal intensity. c: Magnetic resonance cholangio-pancreatography indicates the presence of a large spherical lesion near the confluence of the right and left hepatic ducts that is compressing the intrahepatic bile ducts and keeping them separate from the common bile duct.

Endoscopic retrograde pancreatoc cholangiography. Downward compression of the common bile duct, gall bladder, and cystic duct. No contrast medium is seen in the intrahepatic bile ducts.

Upper abdominal computed tomography. The cystic lesion disappeared after drainage and monoethanolamine oleate treatment.
Figure 6. Upper abdominal contrast computed tomography. Regrowth of the cyst was noted 28 days after the initial drainage, for which a second drainage was performed.

References

12. Matsuzawa K, Inabashi K, Matsuzawa S, et al. A case of large liver cyst with extraordinarily high level of CA19-9 in the serum, vessels, liver damage would likely have continued for several days. As there was no serious liver damage in the present patient, most of the monoethanolamine oleate must have been aspirated. Watanabe et al (20) reported that retrograde infusion of monoethanolamine oleate into the common bile duct of the guinea pig resulted in extensive hepatic necrosis and hyperammonemia. In the present case, no contrast medium was detected in the cystic lesion on endoscopic retrograde pancreatobiliaryography, indicating that there was no communication between the cyst and the common bile duct. This negative finding is very important, because monoethanolamine oleate injection into the liver cyst would present serious risk if it did in fact communicate with the bile duct.

Our patient was diagnosed with a simple cyst based on the basis of imaging and cytology findings. Since neoplastic changes of hepatic cysts have occasionally been reported (21, 22), careful follow-up will be necessary.

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