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CASE REPORT

Percutaneous aspiration and sclerotherapy of a giant simple hepatic cyst causing obstructive jaundice: A case report and review of literature

Xu-Xia He, Mei-Xing Sun, Ke Lv, Jian Cao, Sheng-Yu Zhang, Jing-Nan Li

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Abstract

BACKGROUND
Giant simple hepatic cysts causing intrahepatic duct dilatation and obstructive jaundice are uncommon. A variety of measures with different clinical efficacies and invasiveness have been developed. Nonsurgical management, such as percutaneous aspiration and sclerotherapy, is often applied.

CASE SUMMARY
The case is a 39-year-old female with a 5-mo history of cutaneous and scleral icterus, loss of appetite, and dark urine. Lab tests showed jaundice and liver function abnormalities. Imaging revealed a giant simple hepatic cyst obstructing the intrahepatic bile ducts. A combination of percutaneous catheter aspiration and lauromacrogol sclerotherapy was successfully performed and the effects were satisfactory with the size of cyst decreasing from 13.7 cm × 13.1 cm to 3.0 cm × 3.0 cm. Further literature review presented the challenges of managing giant simple hepatic cysts that cause obstructive jaundice and compared the safety and efficacy of a combination of percutaneous aspiration and lauromacrogol sclerotherapy with other management strategies.

CONCLUSION
Giant simple hepatic cysts can cause obstructive jaundice, and a combination of percutaneous catheter aspiration and sclerotherapy with lauromacrogol are suggested to treat such cases.
Key Words: Simple hepatic cyst; Obstructive jaundice; Aspiration; Sclerotherapy; Lauromacrogol; Case report

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Core Tip: Giant simple hepatic cysts causing obstructive jaundice are uncommon. Here we presented the challenges of managing giant simple hepatic cysts causing obstructive jaundice and compared the safety and efficacy of percutaneous aspiration and lauromacrogol sclerotherapy with other management strategies. The case is a 39-year-old female with jaundice and liver function abnormalities. Images revealed a giant simple hepatic cyst with obstruction of intrahepatic bile ducts. A combination of percutaneous catheter aspiration and lauromacrogol sclerotherapy was conducted successively, achieving satisfactory efficacy. Therefore, a combination of percutaneous aspiration and lauromacrogol sclerotherapy may be suggested to solve such cases.

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INTRODUCTION

Hepatic cysts occur in 2.5%-18% of the population[1-3]. They generally include a cluster of diseases with heterogeneous pathogenesis and etiology, including simple hepatic cysts, infectious cysts, cystic neoplasms, biliary duct-related cysts and some congenital polycystic liver diseases[4]. Most simple cysts are asymptomatic and are incidentally identified during imaging examinations, including ultrasonography (US), computed tomography (CT) or magnetic resonance imaging[5,6]. Only 5%-16% of simple hepatic cysts become symptomatic due to mass effects, rupture, hemorrhaging, or infection[5,7,8]. They mainly present as abdominal pain, nausea, vomiting and occasional jaundice[9,10].

The management of simple hepatic cysts widely differs according to clinical manifestations, imaging features, and, sometimes, patient preference. A watch-and-see strategy is acceptable for asymptomatic simple cysts, whereas interventions are required if cysts cause severe symptoms or complications. Various treatment methods with different clinical efficacies and levels of invasiveness have been developed. For nonsurgical management, percutaneous aspiration, sclerotherapy, and internal drainage are often used[8,9]. Surgical treatment mainly includes unroofing, cyst fenestration, hepatectomy, and open or laparoscopic liver transplantation[11]. Treatment selection depends on cyst location, size, surroundings and other factors[12,13].

Here, we report a case of a giant simple hepatic cyst in the hepatic hilum causing intrahepatic duct dilatation and obstructive jaundice. A combination of percutaneous aspiration and lauromacrogol sclerotherapy was performed and achieved satisfactory effects. The related literature was reviewed to better understand management in similar patients.

CASE PRESENTATION

Chief complaints
A 39-year-old female was admitted for cutaneous and scleral icterus, loss of appetite, and dark urine for 5 mo.

History of present illness
A 39-year-old female was admitted for cutaneous and scleral icterus, loss of appetite, and dark urine for 5 mo.

History of past illness
The patient used to be in good health and had no previous medical history.

Personal and family history
The patient’s personal habits, customs, and family history were unremarkable.
Physical examination
Physical examination revealed moderate jaundice without abdominal tenderness, hepatomegaly, or Murphy’s sign.

Laboratory examinations
Lab tests showed jaundice [total bilirubin (TBil) level was 149.8 μmol/L, and direct bilirubin (DBil) level was 118.7 μmol/L], liver function abnormalities (liver function test levels included the following: Alanine transaminase (ALT) was 175 U/L, aspartate aminotransferase (AST) was 130 U/L, gamma-glutamyl transpeptidase was 454 U/L, alkaline phosphatase was 314 U/L) and moderate anemia [the hemoglobin (HGB) level was 75 g/L]. Tumor markers were unremarkable except for a slightly elevated carcinoma embryonic antigen (CEA) level of 6.1 ng/mL (normal range: 0-5). Antibodies for hepatitis virus, primary biliary cholangitis and autoimmune hepatitis were all within the normal limits.

Imaging examinations
The abdominal US and the endoscopic US showed an enlarged liver (3.7 cm below the xiphoid process) and an anechoic area (increasing from 11.2 cm × 9.9 cm to 13.7 cm × 13.1 cm in three months) with a clear boundary and no peripheral blood flow, and the intrahepatic bile duct of the left lateral segment was approximately 0.6 cm wide. Magnetic resonance cholangiopancreatography showed several hepatic cysts. The largest cyst was approximately 9.5 cm × 11 cm in size, located in the hilum, and obstructed the intrahepatic bile ducts. Three-dimensional reconstruction of the biliary tract showed dilated intrahepatic bile ducts and compressed hepatic vessels and branches of the portal vein (Figure 1).

Notably, esophagogastroduodenoscopy and colonoscopy were performed and excluded gastro-intestinal neoplastic diseases.

FINAL DIAGNOSIS
A giant simple hepatic cyst complicated with obstructive jaundice was the diagnosis.

TREATMENT
We successfully performed a combination of percutaneous catheter aspiration and sclerotherapy with lauromacrogol. During percutaneous catheter aspiration under the guidance of US, the giant cyst was punctured with an 18-gauge pig-tail catheter. Postoperative drainage was favorable, and a total of 800 milliliters of clear yellow fluid was drained; bilirubin levels, tumor markers (such as CEA level) and cytology tests were unremarkable. Jaundice (TBil was 66.4 μmol/L, DBil was 51.2 μmol/L) and liver function anomalies (ALT was 90 U/L, AST was 59 U/L) were significantly relieved soon after drainage.

Then, two sessions of sclerotherapy (lauromacrogol) of the hepatic cyst were performed (30 mL and 20 mL lauromacrogol mixed with triple amounts of air) at one week. Of note, before sclerotherapy, the communications of the cyst with the surrounding bile ducts were ruled out by injecting a diluted contrast medium into the cyst cavity. After sclerotherapy, a small amount of cyst fluid was drained, and the tube was removed. The patient was generally in good condition. He was discharged and experienced further improvement in his liver function (ALT level was 38 U/L, TBil level was 34.9 μmol/L, and DBil level was 33.5 μmol/L; Figure 2).

OUTCOME AND FOLLOW-UP
During follow-up, the patient reported continued resolution of his symptoms. Three months after treatment, the size of the liver cyst decreased to 6.5 cm × 5.6 cm, and liver function returned to normal limits. Fourteen months after treatment, the size of the cyst had decreased to 3.0 cm × 3.0 cm on US.

DISCUSSION
Most simple liver cysts are asymptomatic and stable in size and structure, which allows for observation. However, some of these tumors gradually grow and eventually cause symptoms due to large size, rupture, hemorrhaging, infection, or neoplasm in rare cases[8,14]. Symptoms, including abdominal discomfort or pain, nausea, vomiting, jaundice, early satiety, and even dyspnea[9,10], are largely related to cyst size and location and are more often attributed to larger cysts and right-sided cysts[9,15]. In a recent review, abdominal pain was reported to be the most common symptom of simple hepatic cysts and was reported by 60% (456 of 764) of the patients[16].
Figure 1 Initial radiographic assessments of the cyst. A: Magnetic resonance cholangiopancreatography displayed a giant hepatic cyst approximately 9.5 cm × 11 cm in size located in the hilum and obstructed intrahepatic bile ducts; B-D: Abdominal contrast-enhanced computed tomography and three-dimensional reconstruction of the biliary tract displayed a hepatic cyst 11.0 cm × 10.6 cm × 12.7 cm in size with compressed hepatic arteries and veins and dilated intrahepatic bile ducts. Multiple portal vein branches were also involved, and tortuous vessels were seen around the gastric fundus and the spleen.

Figure 2 Changes in hepatic indicators after treatment. The patient successfully underwent percutaneous catheter aspiration on December 17, 2020, and then two laurumacrogol sclerotherapies on December 21 and December 27. After treatment, liver function continuously dropped to normal limits. ALT: Alanine transaminase; HGB: Hemoglobin; TBil: Total bilirubin; DBil: Direct bilirubin.

Obstructive jaundice caused by solitary simple liver cysts is quite rare. A total of 17 cases of simple or benign liver cysts accompanied by obstructive jaundice were reviewed (Table 1)[17-33]. The average age of the patients was 65.2 years old, with a 7:10 female to male ratio. These cysts tended to be large (greater than 10 cm) and centrally located when compression of the main intrahepatic duct or even the hepatic hilum was present. Treatment for these patients varied from aspiration to resection. In recent years, a combination of drainage, sclerosing agent injection, and deroofing seem to be the most common treatment methods. Choledochoscopy was also proven to effectively treat these patients[33]. In our
## Table 1 Published cases with simple or benign hepatic cysts causing obstructive jaundice

<table>
<thead>
<tr>
<th>No.</th>
<th>Ref.</th>
<th>Age/sex</th>
<th>Cyst (cm)</th>
<th>Location (segments)</th>
<th>Total bilirubin (mg/dL)</th>
<th>Treatment</th>
<th>Prognosis</th>
<th>Follow-up period</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Caravati et al.[17], 1950</td>
<td>33/M</td>
<td>NA</td>
<td>IV, V</td>
<td>NA</td>
<td>Aspiration + marsupialization</td>
<td>Improved</td>
<td>7 mo</td>
</tr>
<tr>
<td>2</td>
<td>Hudson[18], 1963</td>
<td>55/F</td>
<td>25</td>
<td>III, IV, V</td>
<td>14</td>
<td>Cystenterostomy</td>
<td>Improved</td>
<td>1 mo</td>
</tr>
<tr>
<td>3</td>
<td>Dardik et al.[19], 1964</td>
<td>69/F</td>
<td>15</td>
<td>V</td>
<td>9</td>
<td>Cystectomy</td>
<td>Improved</td>
<td>1 mo</td>
</tr>
<tr>
<td>4</td>
<td>Sacks et al.[20], 1967</td>
<td>81/M</td>
<td>20</td>
<td>IV</td>
<td>19</td>
<td>Aspiration</td>
<td>Improved</td>
<td>2 mo</td>
</tr>
<tr>
<td>5</td>
<td>Santman et al.[21], 1977</td>
<td>61/M</td>
<td>15</td>
<td>IV</td>
<td>29</td>
<td>Partial resection</td>
<td>Improved</td>
<td>NA</td>
</tr>
<tr>
<td>6</td>
<td>Machell et al.[22], 1978</td>
<td>67/F</td>
<td>NA</td>
<td>III, IV, V</td>
<td>NA</td>
<td>Drainage + transhepatic T-tube</td>
<td>Improved</td>
<td>7 mo</td>
</tr>
<tr>
<td>7</td>
<td>Morin et al.[23], 1980</td>
<td>80/M</td>
<td>17</td>
<td>IV, V</td>
<td>15</td>
<td>Aspiration only</td>
<td>Improved</td>
<td>10 mo</td>
</tr>
<tr>
<td>8</td>
<td>Fernandez et al.[24], 1984</td>
<td>61/F</td>
<td>30</td>
<td>III, IV, V</td>
<td>22</td>
<td>Partial resection</td>
<td>Improved</td>
<td>24 mo</td>
</tr>
<tr>
<td>9</td>
<td>Clinkcales et al.[25], 1985</td>
<td>80/M</td>
<td>8</td>
<td>IV</td>
<td>8</td>
<td>Aspiration only</td>
<td>Improved</td>
<td>1 mo</td>
</tr>
<tr>
<td>10</td>
<td>Cappel et al.[26], 1988</td>
<td>44/F</td>
<td>12</td>
<td>IV, V</td>
<td>5</td>
<td>Aspiration</td>
<td>Improved</td>
<td>3 mo</td>
</tr>
<tr>
<td>11</td>
<td>Spivey et al.[27], 1990</td>
<td>73/M</td>
<td>11</td>
<td>IV, V</td>
<td>10</td>
<td>Drainage + deroofing</td>
<td>Improved</td>
<td>NA</td>
</tr>
<tr>
<td>12</td>
<td>Terada et al.[28], 1993</td>
<td>71/F</td>
<td>12</td>
<td>III, IV, V</td>
<td>9</td>
<td>Drainage + cystectomy</td>
<td>Improved</td>
<td>1 mo</td>
</tr>
<tr>
<td>13</td>
<td>Yoshihara et al.[29], 1996</td>
<td>88/M</td>
<td>16</td>
<td>IV, V</td>
<td>8</td>
<td>Drainage + minocycline injection</td>
<td>Improved</td>
<td>9 mo</td>
</tr>
<tr>
<td>14</td>
<td>Kanai et al.[30], 1999</td>
<td>71/M</td>
<td>15</td>
<td>IV, V, VIII</td>
<td>5</td>
<td>Drainage + deroofing</td>
<td>Improved</td>
<td>15 mo</td>
</tr>
<tr>
<td>15</td>
<td>Ishikawa et al.[31], 2002</td>
<td>70/M</td>
<td>18</td>
<td>IV, V, VIII</td>
<td>9</td>
<td>Drainage + minocycline injection</td>
<td>Improved</td>
<td>20 mo</td>
</tr>
<tr>
<td>16</td>
<td>Ogawa et al.[32], 2004</td>
<td>64/M</td>
<td>9</td>
<td>NA</td>
<td>NA</td>
<td>Drainage + minocycline injection</td>
<td>Improved</td>
<td>NA</td>
</tr>
<tr>
<td>17</td>
<td>Zhang et al.[33], 2018</td>
<td>41/F</td>
<td>5</td>
<td>IV</td>
<td>24</td>
<td>Choleodochoscopic high-frequency needle-knife electrotomy</td>
<td>Improved</td>
<td>36 mo</td>
</tr>
</tbody>
</table>

NA: Not available.

Patients, the giant liver cyst caused obstructive jaundice and dilatation of the intrahepatic bile duct of the left lateral segment of the liver, which largely accounted for the patient’s symptoms.

Aspiration is generally associated with high recurrence rates[34]. In recent years, percutaneous aspiration combined with sclerotherapy has been widely used as a minimally invasive procedure for simple hepatic cysts with satisfactory results[35-39]. During percutaneous aspiration and sclerotherapy, US- or CT-guided aspiration and drainage are combined with the injection of a sclerosing agent[7,40, 41]. Sclerosing agents with good efficacy include ethanol, iophendylate, tetracycline chloride, doxycycline, minocycline chloride, and hypertonic saline solution[42].

While liquid sclerosing agents may mix with cyst contents and reduce sclerosing effects, foam sclerotherapy was initially used for vascular malformations and has evolved as an alternative for treating simple hepatic cysts[43]. The agents in a foam vehicle can completely destroy the intimal barrier after 2 min of exposure, causing endothelial edema, exfoliation from the tunica media, and thrombogenesis in the tunica media in 30 min[44]. Sclerotherapy using lauromacrogol foam is rarely reported for treating hepatic cysts. In one case report, laparoscopic lauromacrogol sclerotherapy surgery was reported to be safe and effective in patients with IVa, VII and VIII segment simple hepatic cysts, but more studies are needed to confirm their conclusion[45]. Our case report is the first to combine percutaneous aspiration with sclerotherapy using lauromacrogol in treating a giant simple hepatic cyst, thus proving the safety and efficacy of the therapy. Single or multiple sessions of percutaneous aspiration and sclerotherapy for persistent or recurrent symptoms are adaptable based on cyst features, efficacy and doctor or patient preferences.
preference[7]. In our patients, sclerotherapy with lauromacrogol was planned and administered twice to achieve a better sclerosing effect.

Surgical treatment of simple hepatic cysts, such as open or laparoscopic cyst deroofing or hepatectomy, can be effective but may contribute to recurrence and complications[46,47]. Generally, percutaneous aspiration combined with sclerotherapy and laparoscopic deroofing is reasonable for most symptomatic simple hepatic cysts. A systematic review showed that the outcome of percutaneous aspiration and sclerotherapy was excellent, with symptoms that persisted in less than 4% of patients, and both complication and recurrence rates were < 1%[16]. Major complications were reported in 2/265 (0.8%), 6/348 (1.7%) and 3/123 (2.4%), and cyst recurrence rates were 0.0%, 5.6% and 7.7% in patients treated with percutaneous aspiration and sclerotherapy and laparoscopic and open surgery, respectively[16]. Other studies on the advantage of percutaneous aspiration and sclerotherapy compared to surgical techniques reported similar results[13]. These results supported the safety and efficacy of percutaneous aspiration and sclerotherapy in treating symptomatic simple hepatic cysts prior to surgical procedures. Our patient’s outcome suggested that percutaneous aspiration and sclerotherapy could effectively treat simple giant hepatic cysts. Studies concerning cost, hospitalization time, and quality of life are needed to further compare these measures.

CONCLUSION

Giant simple hepatic cysts can obstruct the intrahepatic bile ducts and cause obstructive jaundice. A combination of percutaneous catheter aspiration and sclerotherapy using lauromacrogol can achieve satisfactory results without evident complications compared to surgical interventions.

FOOTNOTES

Author contributions: He XX and Sun MX compiled all relevant information concerning that case and did the literature research; He XX did the drafting and review of the manuscript; Lv K and Cao J did the radiological analysis; Zhang SY did the study concept and design; Zhang SY and Li JN evaluated the whole treatment of the patient and supervised the study; all authors had reviewed and approved the final version of this manuscript.

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