Case Report

Long-term results of percutaneously treated multiple hepatic and splenic hydatid cysts in a pregnant woman

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Abstract
Hydatid disease is caused by Echinococcus granulosus, which is endemic worldwide. This parasitic tapeworm can produce cysts in almost every organ of the body; however, the liver and lungs are the most frequently targeted. 37-year-old multigravida woman with a 10-week pregnancy in whom multiple splenic and liver hydatid cysts were detected by ultrasound. All splenic and liver hydatid cysts were treated percutaneously under US guidance during the 14th week of pregnancy. The catheterization method was used in the treatment of all hydatid cysts. Alcohol was also used as scolicidal and sclerosing agent in all procedures. There were no major complications. A cystobiliary fistula developed in a hydatid cyst treated in the liver. A healthy baby was delivered vaginally at term.

Key words: catheterization: hydatid cyst; pregnancy; interventional radiology.


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Introduction
Hydatid disease is a zoonotic infection caused by Echinococcus granulosus [1]. This infection results in a parasitic disease that is especially endemic in regions [1,2]. The disease most frequently targets the liver and less commonly affects other organs such as lungs, spleen, bones, adrenal gland, kidneys and ovary [3].

Surgical procedures lead to higher morbidity and mortality rates, longer hospital stays, and solid organ losses in the treatment of hydatid cysts [1]. In recent years, however, minimally invasive approaches have been developed, and these methods include percutaneous treatments with excellent results and lower mortality and morbidity rates [4].

In this report, we aimed to present the long-term treatment results of a pregnant woman treated percutaneously for multiple hydatid cysts in the liver and spleen.

Case Report
A 37-year-old woman underwent routine ultrasonography (US) during the 10th week of pregnancy. Incidentally, five and three unilocular cystic lesions with uniform anechoic content with wall formation and mobile internal echogenicities were detected in the liver and spleen, respectively. The patient had no diagnosis of hydatid disease, previously. Laboratory investigations including complete blood count and biochemistry profile were within normal limits. Echinococcal hemagglutination and specific Echinococcus IgE tests were positive. Five cystic echinococcosis (CE) type 1 in the liver and three CE type 1 in the spleen were diagnosed according to laboratory and radiological findings. Cytological investigations of specimens acquired by aspiration confirmed the diagnosis of hydatid cysts during and after the percutaneous treatment.

Percutaneous treatment was chosen because of the risk of rupture of multiple, large volume hydatid cysts. Oral albendazole was started at a dose of 10 mg/kg/day for 7 days at the 13th week of pregnancy before percutaneous treatment and continued at the same dose for 2 weeks after percutaneous treatment to reduce the risk of secondary dissemination. Complete blood count, prothrombin time, partial thromboplastin time, international normalized ratio, and platelet count were evaluated.

The procedure was performed with the patient under conscious sedation and local anesthesia. Propofol (1 mg/kg) and fentanyl (1 µg/kg) were intravenously administered. Subsequent doses were repeated as planned and the patient was monitored by an anesthesiologist in the interventional suite.
All interventional procedures were performed with US guidance. All hepatic and splenic hydatid cysts were individually catheterized in the same operation. After sterilization, the patient was locally anaesthetized with prilocaine hydrochloride and the cysts were punctured through an 8F catheter (Bioteq, Taipei, Taiwan) using a trocar method. All cysts contents were aspirated. For the liver hydatid cysts, lucent cyst contents confirmed that the cyst had no communication with the biliary tract. Absolute alcohol (98% alcohol) was injected into the cysts at a rate of approximately 10% of the aspirated content. After waiting for nearly 10 minutes to visualize detachment of the endocyst from the pericyst on US, cystic contents were re-aspirated. To avoid systemic toxic effects due to the absorption of residual alcohol, the cystic cavity was irrigated with 0.09% NaCl; the catheter was then affixed to the skin.

Post-procedure, the patient was monitored for early complications in the interventional radiology unit and was hospitalized the same day. The following day, all catheters were removed when the amount of fluid drained from the cavity decreased to < 10 mL per day. The catheter of the third liver segment lesion was left in place because drainage did not decrease to < 10 mL, and the patient was diagnosed with a biliary fistula based on the presence of bile-stained drainage fluid. Given the patient’s clinical stability, no other diagnostic or therapeutic procedures were performed. When the drainage fluid lacked bile-staining and the rate of drainage decreased to < 10 mL, the final catheter was removed on the 4th post-procedure day. No major complications occurred.

Follow-up of the patient was performed at months 1, 3, 6, 9, 12, and 24 under US guidance. Table 1 summarizes localization and pre- and post-treatment volumes of the lesions. In all treated cysts, a decrease in cyst volume of more than 50%, membrane detachment, an increase in solid components, and a pseudomotor appearance were identified (Figure 1). The patient delivered a healthy female infant at 38 weeks gestation and no teratogenic effects have been detected during the follow up period.

**Discussion**

Hydatid cysts are rarely seen during the pregnancy, a swift increase may be witnessed in cystic volume due to the decreased immunity during this period [5]. Larger hydatid cysts may rupture in the second half of the pregnancy, and death may ensue [6]. The treatment of pregnant women having a hydatid cyst with a larger size is of vital importance.

Percutaneous treatments are less noninvasive and mortality and morbidity rates are lower [1,7]. Different percutaneous techniques can be performed according to the size and stage of the lesions [3,1,7]. Defined by Akhan et al., the catheterization technique is used to treat cysts larger than 6 cm and requires the placement of a catheter into the cystic cavity [8]. In our patient, the catheterization technique was used under the US guidance; however, different from the catheterization techniques, absolute alcohol was used both as a

**Table 1. Location, pre- and post-treatment volumes, and complications of percutaneously treated hydatid cysts.**

<table>
<thead>
<tr>
<th>No</th>
<th>Location</th>
<th>Pre-treatment volume (cc)</th>
<th>Post-treatment volume (cc)</th>
<th>Reduction rate (%)</th>
<th>Complications</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Liver (Seg 8)</td>
<td>660</td>
<td>90</td>
<td>86.3</td>
<td>None</td>
</tr>
<tr>
<td>2</td>
<td>Liver (Seg 3)</td>
<td>432</td>
<td>46</td>
<td>89.3</td>
<td>Cystobiliary fistula</td>
</tr>
<tr>
<td>3</td>
<td>Liver (Seg 7)</td>
<td>380</td>
<td>35</td>
<td>90.7</td>
<td>None</td>
</tr>
<tr>
<td>4</td>
<td>Liver (Seg 5)</td>
<td>320</td>
<td>41</td>
<td>87.1</td>
<td>None</td>
</tr>
<tr>
<td>5</td>
<td>Liver (Seg 6)</td>
<td>192</td>
<td>12</td>
<td>93.7</td>
<td>None</td>
</tr>
<tr>
<td>6</td>
<td>Spleen</td>
<td>336</td>
<td>99</td>
<td>70.5</td>
<td>None</td>
</tr>
<tr>
<td>7</td>
<td>Spleen</td>
<td>216</td>
<td>14</td>
<td>93.5</td>
<td>None</td>
</tr>
<tr>
<td>8</td>
<td>Spleen</td>
<td>156</td>
<td>5</td>
<td>96.7</td>
<td>None</td>
</tr>
</tbody>
</table>

No: number of lesion; Seg: segment.
scolicidal and a sclerosing endocavitary agent in the same session to treat hydatid cysts. Thanks to the trocar technique, the number of punctures performed in hydatid cyst during the interventions can be reduced; thus, we consider the risks for abdominal spillage and leakage may be avoided.

Albendazole or mebendazole are used in the medical therapy of hydatid cyst, however, these may have teratogenic effects in the first trimester [9]. Several studies have suggested that albendazole be used to prevent the seeding from spillage prior to percutaneous treatments in pregnant women [9]. We administered oral albendazole (10 mg/kg/day) over a period of 3 weeks within the second trimester before and after the interventions due to existing multiple large cysts and to avoid subsequent abdominal dissemination. Our patient delivered a healthy female infant at the 38th week and no teratogenic findings were detected in the newborn. One study has reported that intravenous sedation is a safe modality in the percutaneous treatment of hydatid cysts in pregnant women [9]. In our patient, intravenous sedation and local anesthesia were used as a combined regimen.

Major complications, such as abdominal dissemination and anaphylactic shock, and minor complications, such as urticaria, itching, hypotension, abscess, cystobiliary fistula, hemorrhage and pneumothorax can be seen in treating liver and spleen hydatid cysts percutaneously [3,1]. In addition, pregnancy may accelerate the development of hydatid cysts, and the labor may also be obstructed by hydatid cysts [10]. In our patient, a cystobiliary fistula developed only in the lesion of hydatid cyst treated in the third segment of the liver. This complication did not show much influence on the outcome and was treated with noninvasive techniques.

Conclusion
Hydatid disease is still an important public health problem in endemic countries such as Middle East, Mediterranean basin, South America, Central Asia, Africa, Australia, New Zealand, and Turkey. Percutaneous hydatid cyst treatment has been found safe and effective in many related studies. However, there are very few studies in the literature regarding percutaneous treatment of hydatid cyst during pregnancy. To our knowledge, this is the first case report in the literature presenting the results of percutaneous treatment in a pregnant woman with splenic hydatid cysts. This case report had showed percutaneous treatment of liver and splenic hydatid cysts during pregnancy with the catheterization technique seems safe, reliable, and effective procedure as a serious alternative to surgical interventions. Additional long-term studies with larger patient population are needed.

References

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