Surgical management and long-term outcome of complicated liver hydatid cysts caused by *Echinococcus granulosus*

Roland Chautems, MD,a Leo H. Bühler, MD,a Benjamin Gold, MD,a Emile Giostra, MD,b Pierre Poletti, MD,c Michael Chilcott, MD,c Philippe Morel, MD,a and Gilles Mentha, MD,a

Geneva, Switzerland

**Background.** The aim of this retrospective study was to evaluate clinical presentation and long-term outcome of patients treated surgically for complicated liver hydatid cysts.

**Patients and Methods.** Eighty-four patients with liver hydatid cysts underwent an operation at the Geneva University Hospital between 1980 and 1999. Clinical presentation, postoperative morbidity, mortality, and long-term recurrence rate were evaluated.

**Results.** Among the 84 patients with liver hydatid disease, 35 patients (41%) presented complicated cysts (ie, cysts that had developed a fistula into adjacent structures or organs). In most patients, the fistula communicated with the biliary tree (n = 25), but we also observed communication with the right lung (n = 3), the right diaphragm (n = 2), liver parenchyma (n = 1), and peritoneal cavity (n = 1). Complete removal of the cystic disease was possible in 24 of 35 patients (70%). In 11 patients, fragments of cysts were not removed because of their location adjacent to main vessels. Postoperatively, 8 patients (23%) developed a severe complication (grade II and III). There were no postoperative deaths, and no recurrences of hydatid disease were observed with a median follow-up of 8.6 years (complete follow-up was obtained in 69% of patients).

**Conclusions.** Complicated liver hydatid disease is frequent and was observed in almost half of patients operated for liver hydatid cysts at our center. Using a surgical strategy aimed at complete removal of cystic and pericystic tissue with simultaneous treatment of the fistulous tract, we observed 23% postoperative morbidity, no mortality, and no recurrence of disease with a median follow-up of >8 years. (Surgery 2005;137:312-6.)

From the Department of Surgery, Clinic of Visceral Surgery,a and the Division of Gastroenterology, Departments of Medicine,b and Radiology,c University Hospital of Geneva

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Over time, liver hydatid disease caused by *Echinococcus granulosus* can involve adjacent structures or organs by development of fistulous tracts and dissemination of daughter cysts into the biliary tree, pleura, bronchi, or peritoneum. The incidence of complicated hydatid cysts has been reported to be as high as 60% of patients diagnosed and treated for liver hydatid disease.1

Treatment of liver hydatid disease in general is still a matter of debate, and no clear guidelines are available. It is generally agreed that medical therapy alone is ineffective and unreliable in curing the disease although stabilization has been reported with albendazole alone or in combination with praziquantel2,3

Operative therapies vary from complete resection (eg, total pericystectomy or hepatectomy) to minimal invasive procedures (eg, percutaneous aspiration of cysts).4,6 More recently, reports7-9 have been published on laparoscopic approaches to the treatment of hepatic hydatid cysts. The choice of therapy depends on several factors: general condition of the patient, number and localization of the cysts, surgeon’s expertise, and type of hospital in which the surgery is performed, including the possibility of intensive postoperative care.

Only a few studies10-12 have reported the management and outcome of patients with complicated liver hydatid disease. The aim of our current study was to analyze clinical presentation and the long-term outcome of patients treated operatively for complicated liver hydatid cysts at our center.
PATIENTS AND METHODS

For this retrospective study, we reviewed the charts of 84 patients with liver hydatid cysts caused by *Echinococcus granulosus*, who were treated at our department between January 1980 and December 1999 of which 35 (41%) presented a complicated hydatid cyst. The remaining 49 patients (59%) presented with one or several cysts without invasion of adjacent organs or biliary vessels. Blood tests including complete blood count, liver function tests, and hydatid serology (ELISA and/or hemagglutination test) were obtained. Radiologic workup included ultrasonography and computed tomography. Angiography was performed selectively in patients who had evidence of close contact between a cyst and the main branches of the portal vein or hepatic artery.

Operative strategy was directed toward curative excision of all parasitic tissue and pericystic tissue. Drapes soaked with hypertonic sodium chloride solution (20%) were placed around the cysts before their emptying to produce scolicidal effect. If a possible communication between a cyst and the biliary tree was suspected from the preoperative assessment (eg, cholestasis or dilatation of the biliary tree) or suspected during surgery, intraoperative cholangiography was performed and, if necessary, the biliary tree was explored. Cholecystectomy was not performed routinely.

The charts of the patients were reviewed and the complications classified according to Clavien et al13 (Table I).

Postoperative, antiparasitic treatment with albendazole was given to 13 patients up to 6 months. Only patients with residual cystic fragments, a bronchial fistula, or rupture of a cyst into the peritoneal cavity were considered for antiparasitic treatment. The total daily doses varied between 600 and 800 mg.

Follow-up was organized by our team or by the primary care physician. Follow-up workup included blood tests (same as above) and abdominal ultrasonography on regular intervals. Patients and/or their physicians were sent a questionnaire asking about any recurrence.

RESULTS

Clinical presentation. All 35 patients with complicated liver hydatid disease were born or had lived several years in endemic areas: Algeria (n = 3), Chile (n = 1), Spain (n = 5), Iraq (n = 1), Italy (n = 5), Lebanon (n = 1), Libya (n = 1), Morocco (n = 2), Portugal (n = 7), Switzerland (n = 2), Turkey (n = 3), and Yugoslavia (n = 4). There were 20 females and 15 males aged between 15 to 71 years (median, 40 years). Clinical presentation was variable and included abdominal pain (n = 10), fever (n = 7), jaundice (n = 4), coughing (n = 2), anaphylaxis (n = 2), unexpected discovery (n = 3), no sign or symptom (n = 2) found during follow-up (n = 2); abdominal pain was the most common symptom. Two patients with recurrent hydatid disease had undergone a previous operation at another center. Between 1 and 20 cysts were found per patient. In most patients, a complicated cyst was diagnosed preoperatively (cholestasis or dilatation of the biliary tree), and in only 3/35 patients (8%) was an unexpected fistula discovered intraoperatively.

Description of complicated cysts and surgical therapy. A fistula between a cyst and the biliary tract was detected in 25 patients (30% of all
patients with liver hydatid disease and 71% of those with complicated cysts). The fistula was usually revealed by an open bile duct communicating with the removed cyst and confirmed by cholangiography (Fig 1). At cholangiography, daughter cysts were found located within the common bile duct in 12 of 25 patients (Fig 2). For these patients, daughter cysts were removed by choledochotomy with a T tube left in place for 4 weeks.

After removal of the cyst and the fistulous tract, any open bile duct was closed with a reabsorbable suture. In 1 patient, a cyst was communicating with the bile duct of segment IV, which was distally obstructed by an inflammatory stricture. The patient was treated by a segmental drainage via a Roux-en-Y jejunal limb.

Invasion of the diaphragm was observed in 5 patients (14% of complicated cysts). Surgical treatment consisted of excision of the fistulous tract, local diaphragm excision, direct suture, and pleural drainage.

A fistula into the chest was observed in 3 patients (9% of complicated cysts); 2 fistulas involved the bronchial tree and 1 the pleural space. Associated procedures were excision of the fistulous tract, and lung and pleural drainage.

Preoperative rupture of a cyst into the peritoneal cavity was observed in 1 patient, who presented with acute abdominal pain. Emergency laparotomy revealed a large left liver cyst with free daughter cysts in the peritoneal cavity. A left liver hepatectomy was performed in association with intraoperative peritoneal lavage with 3 L of hypertonic sodium chloride solution. Before closure, the peritoneal cavity was rinsed again with normal saline (154 mmol/L NaCl) solution. This patient was treated postoperatively with albendazole for 6 months.

Intrahepatic rupture of a cyst was found in 1 patient who had undergone a previous operation by pericystectomy for liver hydatid disease. Subtotal pericystectomy and cyst unroofing were performed.

Of the 35 patients with complicated hydatid disease, 24 (69%) had a complete removal of the disease by hepatectomy (12 patients) or total pericystectomy (12 patients). For the remaining 11 patients, some pericystic tissue (subtotal pericystectomy) or part of the cyst itself (cyst unroofing) was left in place (Table II). Only small fragments of cysts adjacent to main vessels (which had to be preserved) persisted after resection. The reason for the incomplete resection was to save main hepatic or portal veins to preserve hepatic function. With regard to surgical treatment of cystic hydatid disease, the risk between disease recurrence and postoperative liver failure was carefully weighed.

### Table II. Surgical procedures of the liver (based on Couinaud’s anatomic classification)

<table>
<thead>
<tr>
<th>Surgical procedure</th>
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<tbody>
<tr>
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</tr>
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</tr>
<tr>
<td>Left hepatectomy (segments II, III)</td>
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</tr>
<tr>
<td>Segmentectomy</td>
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</tr>
<tr>
<td>Total pericystectomy</td>
<td>27</td>
</tr>
<tr>
<td>Subtotal pericystectomy</td>
<td>12</td>
</tr>
<tr>
<td>Unroofing</td>
<td>6</td>
</tr>
<tr>
<td>Cystojejunostomy</td>
<td>1</td>
</tr>
</tbody>
</table>

Fig 2. Preoperative computed tomography scan showing voluminous hydatid cysts in the right and left liver lobes, with (A) a fistula in the right liver extending through the diaphragm (arrow) and into the (B) right lung (arrow). This patient underwent total pericystectomies, unroofing of the cysts, and resection of the fistulous tract in the right lung.
One patient presented with 2 simultaneous lung hydatid cysts and another with a cyst in the retroperitoneum, all of which were removed at the time of the hepatic surgery.

**Postoperative morbidity, mortality, and recurrence rate.** Postoperative complications developed in 13 patients (37%); 8 (23%) of which were classified as severe (grade II and III). Type III complications included a brachial plexus injury in 1 patient after a 10-hour procedure (consisting of unroofing of 4 cysts and 8 total pericystectomies). A post-transfusional hepatitis C developed in 1 patient who received 4 units of blood intraoperatively in 1992. Other complications were treated medically or by drainage. There was no postoperative mortality (ie, all patients were discharged from the hospital in stable condition).

Of the 35 patients, follow-up was complete in 24 (69%) and partial in 11 (31%) with a median of 103 months (15-240) or 8.6 years. No recurrence was found during our study period.

**DISCUSSION**

Complicated hydatid liver cysts have been reported to occur in 15% to 60% of patients with hydatid liver disease at the time of diagnosis.\(^1,10-12\) Specific management and long-term outcome have not been evaluated extensively. Involvement of adjacent structures or organs must be considered in preoperative planning.

In our series of 84 patients undergoing an operation for liver hydatid disease, almost half (41%) presented with complicated hydatid liver disease. Most frequently, a communication with the biliary tree was observed (30%). This complication has been reported in the literature to occur in 9% to 30% of patients.\(^1,10-12\) The high rate observed at our center could be explained by referral of patients from non-endemic areas, where delayed diagnosis is more frequent. The fistula was large enough to allow daughter cysts and debris to migrate into the biliary tree in almost half of the patients with biliary fistulas and in 14% of all patients with hydatid liver disease. We recommend performing an intraoperative cholangiography in all patients who present with abnormal liver function tests preoperatively, bile-stained cyst fluid, or intraoperative bile leak after a pericystectomy.

Several authors have advocated pre- or postoperative endoscopic retrograde cholangiopancreatography (ECRP).\(^14,16\) If performed in conjunction with a sphincterotomy, an ECRP may be used to drain an infected biliary tree or to clear the biliary tree before a planned procedure, thereby avoiding the need for an intraoperative cholangiography and bile duct exploration. The reports of such series involved a limited number of patients (n = 11-14), and the success rate was variable (43%-100%).\(^14,16\) At our center, intraoperative cholangiography with or without subsequent duct exploration remains the method of choice. Using this technique, no postoperative biliary obstruction occurred. However, ERCP and sphincterotomy might play a role in patients with acute cholangitis or postoperative biliary complications.

Cysts located at the periphery and at the dome of the liver tend to involve the diaphragm and often the intrathoracic structure. For these patients, local diaphragm excision and suture appeared satisfactory in our experience. Rarely, cysts either rupture into the pleural space or into the bronchial tree.\(^10-12,17\) Surgical management of such complicated cysts can be performed by laparotomy through the diaphragm or by thoracotomy with removal of the involved lung tissue, bronchial suture, and pleural drainage.\(^17\)

Rupture of a cyst into the peritoneal cavity should be a rare event that can be caused by direct trauma, with the risk of an anaphylactic shock. Profuse peritoneal lavage with hypertonic sodium chloride appears to be mandatory to prevent intra-abdominal recurrence of hydatid disease.

Postoperative complications involved 37% of the patients presenting complicated hydatid liver disease. Recently, Gollackner et al\(^18\) reported a series of 74 patients treated for liver hydatid cysts. Postoperative morbidity and mortality were 24% and 3%, respectively.\(^18\) The higher complication rate observed in our report might be explained by the selection of patients presenting with complicated hydatid disease, necessitating more complex surgical procedures. In other series, overall postoperative morbidity was only 3% to 12%.\(^11,19\) However, only complications directly related to the surgical procedures were reported; this difference emphasizes the importance of using an internationally accepted classification to analyze and report postoperative complications.

We have recently reported a higher recurrence rate of disease in patients undergoing an incomplete resection compared to those undergoing complete surgical resection of liver hydatid disease (ie, 12% vs 0%, respectively).\(^20\) Recurrence is likely due to residual vesicles left in place, even if the cysts were carefully emptied. Vesicles can develop from a main cyst and grow next to it, which is especially the case in older cysts. Recurrences may also develop if peritoneal soiling occurs during emptying of a cyst. Recurrence usually becomes
symptomatic only 3 to 4 years after the operation. In the present study, despite the observation that some patients with complicated liver hydatid disease had incomplete surgical resection, no recurrences have been observed to date in the 11 patients who underwent incomplete pericystectomy of all cysts. One reason might be that only a small fragment of cyst(s) adjacent to preserved main vessels persisted after the resection.

CONCLUSION

We recommend complete resection of hepatic hydatid lesions with simultaneous treatment of fistulous tracts located in adjacent structures or organs whenever possible. This strategy allowed us to achieve a low recurrence rate with no mortality and a morbidity of 23%.

REFERENCES