

# Long term results after complete or incomplete surgical resection of liver hydatid disease

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## Summary

**Background:** The liver is the organ most frequently infected by hydatid disease and medical therapy alone is ineffective in eliminating the parasite. Surgical options vary from complete resection (e.g. total pericystectomy or hepatectomy) to limited procedures (e.g. percutaneous aspiration or unroofing of cysts). The aim of this study was to determine the long-term outcome after complete or partial resection of liver hydatid cysts.

**Patients and results:** Between 1980 and 1996, 78 patients were operated upon at our institution for liver hydatid cysts. In Group 1, complete resection was achieved in 57 patients (73%), whereas in Group 2, incomplete resection was performed in 21 patients (27%), due to multiplicity, bilaterality of cysts or close contact between a cyst and por-

tal or hepatic veins. The post-operative morbidity in Groups 1 and 2, was 31% and 47% (N.S.), respectively. Mean duration of hospital stay was 17 and 26 days ( $p = 0.004$ ), respectively. Recurrence rate of hydatid disease after a mean follow-up of 6.6 years was 0% and 12% (N.S.), respectively. There was no mortality in either group.

**Conclusions:** Complete surgical resection of hepatic hydatid disease should be attempted whenever possible. Our results, with a mean follow-up of 6.6 years, indicate limited post-operative debilitating complications, low recurrence rate and no mortality.

**Key words:** *hydatidosis; liver; Echinococcus granulosus; surgical resection*

## Introduction

Hydatidosis is an endemic parasitic disease in countries where sheep are prolific. This is particularly the case in Mediterranean countries including North Africa, Spain and Portugal [1]. The parasite, *Echinococcus granulosus*, is a cestode that grows in the small intestine of its definitive host, usually a dog. Eggs are eliminated in the faeces and when ingested, liberate their larvae in the duodenum of an intermediate host (sheep or humans). The larvae cross the intestinal wall and via the portal system reach the liver where they form cysts. The liver is the organ most frequently infected by this disease (50–93%) [2–4].

Without treatment, cysts grow and eventually form fistulas into adjacent organs or rupture into the peritoneal cavity requiring emergency surgery. Older cysts have an increased risk of exogenous daughter cyst formation, which is an important factor for recurrence of disease after surgery [5–8].

Different therapies have been suggested. Medical therapy alone is insufficient to cure the disease, although stabilization has been reported with albendazole alone or in combination with praziquantel [9–12].

Surgical therapy varies from complete resection (e.g. total pericystectomy or hepatectomy) to minimal invasive procedures (e.g. percutaneous aspiration of cysts) [7, 13–17]. More recently, reports have been published on laparoscopic surgery for hepatic hydatid cysts [18–24]. The choice of therapy depends on several factors: general condition of the patient, number and localisation of the cysts, the surgeon's expertise and the type of hospital where such surgery is performed, including the possibility of intensive post-operative care.

This study presents the long term results in patients undergoing complete or incomplete surgical resection of liver hydatid cysts.

## Methods

### Patients

Between 1980 and 1996, 78 patients were operated upon for liver hydatid cysts at our institution. The study group consisted of 48 women (62%) and 30 men (38%). Age ranged from 15 to 73 years (mean age 40.4 years). Most patients came from countries where parasitic infection is endemic. Seven out of 8 Swiss patients were born or had travelled in these countries (table 1).

### Diagnosis

For 44 patients (56%), abdominal pain was the first symptom leading to the diagnosis of hydatidosis, whereas in 20 cases (26%) the diagnosis was made incidentally during a medical check-up. Nine patients (12%) had an abrupt presentation of the disease following the rupture of a cyst. These patients presented with a hypersensitivity reaction (pruritus, urticaria, anaphylaxis), fever of sudden onset, acute abdominal pain or vomiting of pieces of cysts (table 2). One patient was diagnosed with a hydatid cyst peroperatively during an elective procedure for peptic ulcer disease. Eight patients had a recurrence of their disease between 3 and 35 years after one or more operations in

their home country. The type of initial operation performed was not known exactly, but in most cases this was essentially minimal surgery (emptying, marsupialization, or unroofing of cysts).

### Pre-operative evaluation

Pre-operative evaluation included blood tests (complete blood count, liver function tests and anti-echinococcus antibody testing) and radiological imaging. Abdominal echography and computerised tomography was done routinely. If vascular compression was suspected, angiography or porto-caval venography was performed.

### Surgery

Group 1: Complete resection of the cyst(s) was performed whenever possible, i.e. by total pericystectomy or hepatectomy. Group 2: When complete resection of a cyst was impossible due to multiplicity, bilaterality or close contact between cyst and portal or hepatic veins, unroofing was performed. The latter technique involved placing towels soaked with a hypertonic saline solution around the cyst, emptying it with an aspiration cannula and finally opening and removing of the roof. No scolical agent was injected under pressure into a cyst at any time.

If the pre-operative assessment suggested a possible communication between a cyst and the biliary tree (e.g. cholestasis or dilatation of the biliary tree) or this was suspected during surgery, peroperative cholangiography was performed and, if necessary, the biliary tree was explored. Cholecystectomy was not performed routinely.

### Follow-up

After surgery, patients were followed up, either by our department or by a primary care physician. A complete follow-up was achieved in 48 cases (62%), whereas 23 patients (30%) had an incomplete follow-up and 7 patients (8%) returned to their native countries after the operation with no follow-up. Patients who had a radical resection underwent abdominal echography or computerised tomography one year after the operation. Those patients with less definitive surgery, however, underwent radiological imaging every six months.

### Data analysis

Complication rates according to the type of surgery, mean hospital stay and recurrence rate of hydatid disease were analysed. Statistical analysis was calculated using  $\chi^2$  square and Student test.

**Table 1**

Native country and corresponding number of patients.

Portugal: 15	Ex-Yougoslavia: 4	Chile: 1
Italy: 13	Morocco: 3	Uruguay: 1
Spain: 12	Libya: 3	Greece: 1
Switzerland: 8	Tunisia: 2	Great-Britain: 1
Turkey: 5	Lebanon: 2	France: 1
Algeria: 5	Ethiopia: 1	

**Table 2**

Symptoms leading to discovery of hydatid disease.

Abdominal pain	44 (56%)
Fortuitous discovery	20 (26%)
Pruritus or urticaria	3 (4%)
Emesis	2 (3%)
Anaphylactic shock	1 (~1%)
Traumatic rupture	1 (~1%)
Abdominal mass	1 (~1%)
Fever	1 (~1%)
Pneumonia	1 (~1%)
Thoracic pain	1 (~1%)
Nausea	1 (~1%)

## Results

### Operations

The different types of surgical techniques are summarized in table 3. Patients were divided into 2 groups. Group 1: surgery was considered curative with complete resection of the cyst (s) in 57 patients (73%). Group 2: incomplete resection of cysts was performed in 21 patients (27%).

Thirty-five patients from both groups (45%) had complicated cysts, which communicated with or had perforated into other organs. Communication between a cyst and the biliary tree was found in 27 (35%), with the right lung through the di-

aphragm in three (4%), with the right diaphragm in two (2%), with liver parenchyma in one (1%) and with the peritoneal cavity in one case (1%).

### Adjuvant medical therapy

Thirteen patients (16%) with complicated cysts received peri- and/or post-operative albendazole therapy. Duration of albendazole treatment ranged from a few days to 6 months depending on side effects of the medication and compliance of the patient. The total daily doses varied between 600 and 800 mg.

**Table 3**

Types of surgical techniques performed in 78 patients.

Group 1: complete resection (57 patients)		group 2: subtotal resection (21 patients)	
Total pericystectomies	60	subtotal pericystectomies	20
Right hepatectomies	10	unroofing of cysts	8
Right lobectomy	1	cysto-jejunostomy	1
Left hepatectomies	4		
Left lobectomies	8		
Segmentectomies	8		

**Table 4**

Post-operative morbidity.

Types of complications	group 1 (nbr. of patients)	group 2 (nbr. of patients)
I	12	1
II a	5	1
II b	1	5
III	0	3
IV	0	0

Classification by Clavien et al. [25]

Grade I:	alterations from the ideal postoperative course, non-life-threatening and with no lasting disability. Complications of this grade necessitate only bedside procedures and do not significantly extend hospital stay.
Grade II:	complications which are potentially life-threatening but without residual disability. A subdivision is made according to the requirement for invasive procedures (IIb).
Grade III:	complications resulting in residual long-term disability, including organ resection or persistence of life-threatening conditions.
Grade IV:	complications leading to patient death

### Complications

In group 1, 18 out of 57 patients (32%) developed post-operative complications, compared to 10 among 21 patients (48%) in group 2 (N.S.). The complications were classified according to Clavien et al. [25] from grade I to grade IV (table 4).

Grade I, IIa, and IIb complications consisted of pleural effusions ( $n = 10$ ), parietal infections ( $n = 5$ ), biliary fistulae ( $n = 3$ ) and a sub-diaphragmatic collection ( $n = 1$ ). These problems resolved spontaneously or by simple drainage in all but one patient, who required relaparotomy to treat active haemorrhage of the phrenic artery.

Grade III complications (complications resulting in a prolonged or definitive disability) were:

- injury to the right brachial nerve plexus (with incomplete sensory-motor recovery) in 1 patient due to positioning on the operating table.
- development of hepatitis C in two patients, possibly due to transfusion of infected blood.

The mortality rate (grade IV complications) was 0.

### Duration of hospital stay

Mean hospital stay of Group 1 and 2, was 17 (6–53) and 26 days (10–77), respectively ( $p = 0.004$ ).

### Recurrence

After a mean follow-up period of 79 months (4–207) or 6.6 years, two patients from Group 2 initially operated at our institution developed disease recurrence. Therefore, recurrence rate of Group 1 and 2, was 0 and 12% (N.S.), respectively.

- In one case, recurrence occurred 3 years after subtotal pericystectomy. Recurrence was revealed by ultrasound during a regular follow-up. Initially, a cyst could not be completely removed due its close contact with a sub-hepatic vein. The patient was reoperated on with unroofing of the cyst and has had no further recurrence in 17 years.
- In the second case, recurrence occurred 4 years after subtotal pericystectomy. The patient was lost to follow-up after the first operation and was readmitted emergently to hospital when a recurrent cyst perforated into liver parenchyma, requiring a right hepatectomy.

## Discussion

Our results suggest that a radical surgical approach (i.e. hepatectomy, total pericystectomy) of liver hydatid disease offers better results in terms of morbidity, duration of hospital stay and recurrence rate, compared to incomplete removal of parasitic lesions.

Less invasive procedures (i.e. percutaneous or surgical drainage of the cysts) are still therapeutic options, but patients treated with these methods need close and long-term follow-up. Recurrence

of disease is likely even years later, as has been confirmed by other centres [5, 8, 26, 27].

### Surgery and complications

Over 30% of our patients developed post-operative complications. The majority were minor problems without long-term disability. Only 3 patients (4%) suffered from long-lasting disabilities related to surgery (grade III). Complication rate was lower in Group 1 compared to Group 2 (32 vs.

47%), however this difference was not significant. Overall bile leakage rate was 4%, which is similar to results published by other centres (3–6%). With more conservative approaches, however, this rate may reach 14 to 27% [27]. In our study, no post-operative deaths were observed in either group, indicating that complete removal of parasitic lesions by radical surgery can be achieved without increased mortality.

### Complicated cysts

Almost half of our patients (45%) had complicated hydatid cysts, which is similar to other reported series (36–40%) [2, 26]. Most of the fistulae were with the biliary tree and were diagnosed during surgery.

### Peri-operative use of scolical agents

Drapes soaked with hypertonic saline solution (at least a 20% solution to have scolical effect) were placed around the cysts before their emptying, in order to prevent any soiling of the peritoneal cavity with contents of the cysts [33]. Caution must accompany peri-operative use of scolical agents. Several authors reported complications following the use of formaldehyde solution, including the development of sclerosing cholangitis [26]. Therefore, we injected no scolical agents into cysts during surgery in order to avoid any contact between the agent and the biliary tree in case of biliary fistula. It appears that the risk of sclerosing cholangitis is related to the presence of a communication between a cyst and the biliary tree, to the duration of exposure to the scolical agent or a particular sensitivity to this agent [28–32]. Furthermore, use of peri-operative scolical agents has not yet been clearly proven to be useful.

### Duration of hospital stay

Mean hospital stay was shorter in patients undergoing radical surgery compared to patients treated by partial removal of parasitic lesions (17 vs. 26 days,  $p = 0.004$ ). This difference can be explained by a lower complication rate, but it must also be mentioned that drainage procedures were more widely used in Group 2. Drainage was related to longer hospital stay (23 vs. 15 days,  $p = 0.004$ ), which has also been reported by other centres [34].

### Adjuvant medical therapy

Albendazole has been reported to have some scolical effects [35]. Pre-operative use of this

drug aims to reduce viability of cysts [36, 37] and may be of benefit in reducing risk of spillage in the event of rupture of a cyst during surgery.

After surgery, the recommended daily dose of albendazole is 800 mg [35]. Continuous therapy appears to be more effective than repeated cycles and should last at least 6 months [10]. This treatment seems to reduce the risk of recurrence [37]. For these reasons, we treated patients with complicated cysts before and after surgery for approximately 6 months.

### Recurrence

Overall recurrence rate was 3% with a mean follow-up period of over 6 years. Both recurrences occurred in patients of Group 2 (subtotal pericystectomy). Therefore, risk of recurrence seems to be increased after incomplete resection compared to complete removal, although the difference was not significant (12% vs. 0%, N.S.). This result is confirmed by data from the literature [26, 38–42]. Recurrence is likely to be due to residual vesicles left in place, even if 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 will become symptomatic 3–4 years after surgery [38, 43–46]. Several studies advocating minimally invasive approaches do not mention any follow-up or report short follow-up periods that do not properly assess success of treatment [13, 14, 45, 47].

### Conclusions

We recommend complete resection of hepatic hydatid lesions, provided that localisation of the cysts is favourable and that the technical and logistic infra-structure is available. This study demonstrates that this approach can result in a shorter hospital stay, limited post-operative debilitating complications, low recurrence rate and no mortality.

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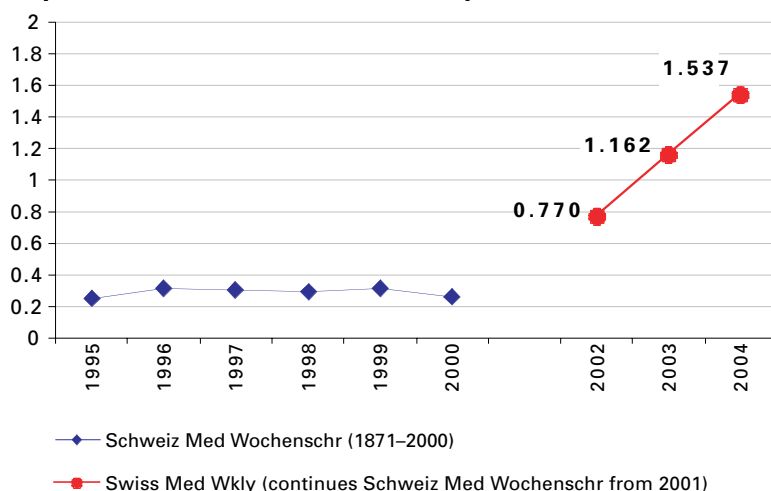
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