# Outcome of Biliary Fistula After Liver Hydatid Cyst Surgery

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## Abstract

**Background:** Biliary fistula after liver hydatid cyst surgery was occur in about 17.8 percent of patients in our center.

**Objective:** To study the outcome of biliary fistula regarding factors related to its causes and management.

**Patients and Methodology**: This is a prospective study of 241cases submitted to liver hydatid surgery in Al-Sader teaching hospital in Al-Najaf city, from October 2015 to April 2019. A total of 43 patients who underwent surgery complicated with persistent leakage of bile from externaldrain. Prospectively collected database were gathered from patients medical records,sent for essential investigations, and treated either conservatively to give a chance for spontaneous closure, or by intervention with ERCP.

**Results:** Forty three patients who develop postoperative biliary fistula after hydatid cyst surgery from 241 patients(17,8 %),230 patients treated with conservative surgery (95,4 %), 2 patients treated with radical surgery(0,82 %) and 9 patients treated with percutaneous aspiration injection reaspiration (3.7%).All the 43 patients in this search treated with conservative surgery. Size more than 15 cm is a risk factors. Intraoperative risk factor is mainly presence of cystobiliary communication which occur in 16 patients (37,2%). Treatment;is conservative in about 23 patients(53.4%).Treatment with ERCP in 19 patients (44.1%).Time of intervention is from 9 – 15 days.

**Conclusion**: Most of patient with external biliary fistula after hydatid liver surgery can be treated conservatively and resolve spontaneously specially low output fistula.

***Keywords:*** *Hydatid cyst, biliary fistulae, Outcome, surgery.*

## Introduction

Hydatid disease is a zoonosis, caused by tapeworm Echinococcus infestation in its larval or cyst stage. The dogs transmit the diseaseto the Humans, and there is no human-to-human transmission. Three known forms of echinococcosis in humans; E. granulosus, E.multilocularis and E. vogeli.[1,2]

The typical hydatid cyst has a three layer wall containing a fluid. The outer one is the **pericyst**, the outer layer of the cyst itself is the laminated membrane (**ectocyst**) and the inner one (**endocyst)** is the germinal membrane, responsible for the production of clear fluid.[3]

**Clinical Features:** Mostly asymptomatic until complications occur. The commonest presentations: dyspepsia,abdominal pain and vomiting. Most common sign is hepatomegaly. The commonest complication is intrabiliary rupture of hydatidcysts,or may rupture to bronchial tree, or free rupture into cavities(peritoneal,pleural,or pericardial).Free ruptures can result in a potentially fatal anaphylactic reaction or disseminated echinococcosis.[3]

**Diagnosis:** The diagnosis of hydatid disease is based on an immunological test for echinococcal antigens [enzyme-linked immune sorbent assay (ELISA)], in approximately 85% of patients is positive. In approximately 30% of patients, Eosinophilia is seen. For detecting hydatid cysts, abdominal Ultrasonography

and CT scanning are both sensitive. Abdominal MRI may be helpful to asses the characteristics features of pericyst, cyst matrix, and daughter cyst.[4]

**Treatment:** Medicl with benzimidazole compounds is currently indicated in

(a) primary hepatic and pulmonary cystic echinococcosis in inoperable patients.

(b) cysts in two or more organs.

(c) small multiple (5 cm) hepatic cysts.

(d) deep cysts in hepatic parenchyma .

(e) secondary prevention .

(f) recurrent.

(g) unfit elderly with unilocular cysts.

(h) specific sites (such as brain, bone, eye)

**Contraindicated for:**

(a) large cysts (10 cm).

(b) honeycomb cysts.

(c) superficial cysts that are prone to rupture.

(d) infected one.

(e) inactive one.

(f) calcified cysts and asymptomatic.

(g) chronic hepatic disease(sever).

(h) bone marrow depression.

(i) pregnancy(early).

(j) Diabetes is a relative contraindication.[5]

**Minimally Invasive Techniques:**

**Percutaneous Drainage of Hydatid Cysts (PAIR)** indicated for:

(a) inoperable patients,

(b) patients who refuse surgery,

(c) Gharbi type I and II,

(d) relapse after surgery,

(e) infected cysts,

(f) failure of chemotherapy,

(g) cysts of greater than 5-cm diameter and in different liver segments,

(h) possibly pregnant women (chemotherapy contraindicated).

(i) possibly children less than 3 years old.

**Contraindications for PAIR are:**

(a) inaccessible cysts,

(b) superficial cysts,

(c) honeycomb cysts,

(d) cysts show hyperechogenic solid patterns,

(e) cysts communicating with bile ducts,

(f) partially or totally calcified cysts,

(g) cysts in the lung.

**Surgery:** Surgerical intervention in the management of liver hydatid is still the "gold standard".

**Indications for open surgery are:**

(a) large one with multiple daughter cysts,

(b) superficial cysts that may rupture,

(c) infected cysts,

(d) cystobiliary communication,

(e) cysts with pressure effect on adjacent organs,

**Contraindications for surgery are:**

(a) patients refusing surgery,

(b) extreme age,

(c) pregnant women,

(d) concomitant severe diseases,

(e) numerous cysts,

(f) difficult to access,

(g) dead cysts,

(h) calcified cysts,

(i) small cysts <5 cm (wait and see policy).[5]

**Postoperative Complications:** The incidence and significance of postoperative morbidity after conservative procedures has been exaggerated in many reports. The most frequent postoperative complications are:

(a) wound infection,

(b) chest problems,

(c) subphrenic abscess,

(d) biliary leaks, and

(e) liver abscess.

**Biliary leak and fistula:**

**Biliary fistula** is bile flow along an abnormal epithelial connection from bile duct into nearby hollow structure, it is either classified as

- **External** or **internal**, external mean connection to the abdominal wall through the wound of its surgery, internal type is the connection to small bowel (bilioenteric) or pleural space (thoracobiliary) or classified as
- **Primary** or **secondary**, primary type which caused by biliary lethiasis or neoplasia,secondary type caused by iatrogenic injury during cholecystectomy or other biliary surgry.[6]

**Biliary leak** is bile escape the bile duct through a perforation e.g rupture hydatid cyst (internal type) or faulty surgical anastomosis . In postoperative hydatid cyst surgery, bile leakage is drainage of bile through abdominal drains, regardless of quantity, no longer than 10 postoperative days. Persistent, stable external drainage of bile for 10 days or more,is a biliary fistula.

The most frequent causes are

(a) An overlooked cystobiliary communication,

(b) Injury to a bile duct,

(c) Inadequately managed cystobiliary communication,

(d) Calcified pericyst,

(e) Hydatid debris obstructing the CBD.[7]

**Patients and Method**

This is a prospective study of 241 cases submitted to liver hydatid surgery in Al-Sader teaching hospital in Al-Najaf city, from October 2015to April 2019. Postoperatively,43 of those patients develop biliary fistula.

Prospectively collected database were gathered from patients medical records including: personal data, medical history of disease, main symptoms and sign, radiological finding (site and size of cyst, single or multiple, primary or secondary, other organs involvement), and ultrasonographic findings, CXR, preoperative complications (dilated biliary tree, jaundice,cyst rupture), type of surgical intervention, presence or absence of cystobiliary communication at operation time and type of its treatment, postoperative follow-up, daily fistula output, time of spontaneous closure, time of intervention, hospital stay postoperatively and mortality.

A biliary fistula is heralded by early postoperative external drainage of large quantities of bile.

Investigations that done to the patients include: liver function test, blood picture, ultrasonographic examination of an abdomen, computerized tomography (not used routinely).

Treatment of patients with biliary fistula includes either conservative management to give a chance for spontaneous closure, or by intervention with ERCP and sphincterotomy with or without stent.

## Results

241 patients with hydatid cyst of liver who undergoes different surgical modalities include;

1. Conservative procedures by deroofing and omental patch in 230 patients (95.4 %).
2. Radical procedures by radical cystectomy and liver resection in 2 cases (0.80 %).
3. Percutaneous aspiration injection reaspiration (PAIR) done in 9 patients out of 241 patients (3.73%).

There are 43 patients out of 241 patients were developed external biliary fistula (17.8 %) subjected to different types of management either surgically or conservatively (Table I):

**Table 1. Characteristics of 43 cases with postoperative biliary fistula after liver hydatidsurgy.**

| **Variable** | | |
|---|---|---|
| Age (Year) | Mean ± SD* | 36.4±7.6 |
| | Range | 18-55 |
| Gender | Female n (%) | 24(55.8 %) |
| | Male | 19(44.1 %) |
| Hospital stay (days) | | 7-19 |
| Mortality** | | 1(2.31 %) |

*SD: standard deviation, **died because of myocardial infarction.

Concerning the location and features of liverhydatid,can summarized in Table II.

**Table 2. Features of the liver hydatid cyst in the patient with biliary fistula**

| Cyst characters | Number | Percentage |
|---|---|---|
| Right lobe | 27 | 62.79 % |
| Left lobe | 12 | 27.9 % |
| Bilobar | 4 | 9.30 % |
| Single | 29 | 67.4 % |
| Multiple | 14 | 32.5 % |
| Primary | 36 | 79.06 % |
| Recurrent | 7 | 16.2 % |
| High output >300 ml/day | 9 | 20.93 % |
| Low output <300 ml/day | 34 | 79,06 % |
| Cystobiliary communication | 16 | 37.2 % |

**Table 3. Hydatid cyst size related to postoperative biliary fistula:**

| Diameter | Number of fistula | Percentage |
|---|---|---|
| 7-10 cm | 6 | 13.9 % |
| 10-15 cm | 11 | 25.5 % |
| 15-22 cm | 26 | 60.4 % |

The output of fistula, the closure time, and Management modality, time and type of intervention,can summarized in Table IV,

**Table 4. Features of patients with biliary fistula:**

| Character | Range |
|---|---|
| Fistula closure time spontaneously in low output fistula<300ml | 19-40 day |
| Fistula closure time in high output fistula | 19- 117 day |
| Fistula output ml/day | 190-810 ml/day |
| Time of intervention in low output fistula no reduction in amount * | 21-28 |
| Time of intervention in high output fistula * | 9 – 15 |
| Drain removal | 21-119 day |

*by ERCP and sphincterotomy with or without stent.

**Table 5. Management of post-operative patients with biliary fistula;**

| Type of treatment | | Number | Percentage |
|---|---|---|---|
| Conservative (spontaneous closure) external drainage | | 23 | 53.48% |
| Endoscopic | ERCP sphincterotomy | 19 | 44.18 % |
| Reoperative | Hepatico-enterostomy | 1 | 2.32 % |
| | CBD exploration+t.tube | 0 | - |

Intra-operatively,the bile leak was discovered in 16 patients, managed by different modalities as in Table VI.

**Table 6. Managements performed for 16 patients with intraoperative bile leak**

| Management | No. of patients | % of patients |
|---|---|---|
| Suturing of cysto-biliary communication with external drainage | 6 | 37,5 |
| Only external drainage | 3 | 18,75 |
| Cyst evacuation +capitonnage | 5 | 31,25 |
| Evacuation with omentoplasty | 2 | 12,5 |

## Discussion

Surgery play an important role in treatment of liver hydatid. Operative options include radical surgery or conservative surgery as the patients in current study were treated. In this study there are 43 patients whose develop biliary fistula after surgery.

Female more common than male, 24 (55.8%) for female and 19 (44.18%) for male, in other research we found (73.2 %) for female and (26.8 %) for male, this is may be related to dealing with vegetable more regarding female side[8], age incidence is ranged from 18 to 55 years old with mean age (36.4 ± 7.6). In other research it was found similar range,Yang X et al [9].

Intraoperative cystobiliary communication was found in 16 patients (37.2%), only 6 of them treated with direct suturing and omental patch before they develop biliary fistula, similar percentage in other research was found; Demircan O et al[10,11]. In comparison with other study; Bhattarai et al, about 13 patients from thirty patients were found with intraoperative cystobiliarycommunication which is close percentage[7,10].

A correlation between biliary fistula and site(right, left or both) in this research as it is more common in right lobe in 27 patients (62.79%) than the left lobe with 12 patients(27.9%) as it found in a similar research Gokhan et al[12]. Kayaalp et al. of 113 patients showed that the site of the liver hydatid close to the hilum is a risk factor for a cystobiliary communication[13].

The cyst size was recognized as a predictive factor in the biliary fistula development. In literature, suggested size more than 10 cm plays a role in a cystobiliary communication development[14], compared to our study it found that the great percentage is when the cyst is more than 15 cm which was in 26 patients (60.4 %).

Multiple studies reported a history of cholangitis, high bilirubin and ALP level,a cyst size more than 10 cm and the presence of suggestive radiological findings as clinical predictors of intrabiliary rupture, and an ERCP was indicated in these cases to delineate the cyst-biliary communication[15].

In our study,spontaneous closure without any intervention was occur in 23 patients of 43(53.48%) in time ranged between (19-35) days,all of them with low output fistula.

In Balik AA et al a review of 304 cases,spontaneous closure in time ranged between 2-4 months inall the 10 cases with external biliary fistulae[16]. In other studies,7 of 12 fistulae closed spontaneously, with the closure time up to 38 days[17]. In this study the closure time of fistula was ranged from 19 to 40 days in cases of low output fistula with gradual reduction in the amount of bile in the drain which was in 23 patients, in cases of high output fistula, closure time was ranged from 19 to117 day with intervention. Time of intervention in low output fistula with no reduction in amount ranged form 19-28 days in other study.

Skroubis G, et al it was found that ERCP was used for the management of high-output fistulas of more than 1 week's duration without reduction and low-output fistulas of more than 3 weeks' duration without reduction of leakage[18].

19 patients in the other hand have been treated by ERCP, they have high output fistula, 2 cases of these 19 patients submitted to more than one ERCP and bigger size stent, in this search the time to intervene is ranged from 9-15 days.

In other cases when there are low output fistula but there is no decrease in amount of output, so intervention is mandatory and not to delay more than 28 days. The time from intervention to closure of fistula also ranged from 10 to 101 days.

## Conclusions

Conservative treatment is the first method of treatment in low output fistula. High output fistula better not to delay intervention and treated by ERCP and sphincterotomy with or without stenting.

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**Conflict of Interest:** None to declare.

**Ethical Clearance:** All experimental protocols were approved under the Dept. of surgery and all experiments were carried out in accordance with approved guidelines.

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