Journal of Surgery
Maaroufi, FK, et al. J Surg 10: 11518
www.doi.org/10.29011/2575-9760.11518
www.gavinpublishers.com
Case Series

Ruptured Hepatic Hydatid Cyst into the Bile Ducts: Case Series Study

Fathillah el Karim Maaroufi[1], Soumaya Jamil[1,2*], Mohamed Boulatar[1], Wafae Ait Belaid[1], Tariq Ahbala[1], Khalid Rabbani[1], Abdelouahed Louzi[1]

[1]Department of General Surgery, Arrazi Hospital, Mohamed VI University Hospital Center, Marrakech, Morocco

[2]Faculty of medicine and pharmacy , Cadi Ayyad University , Marrakech, Morocco

***Corresponding author:** Soumaya Jamil, Department of General Surgery, Arrazi Hospital, Mohamed VI University Hospital Center, Marrakech, Morocco

Citation: Maaroufi FK, Jamil S, Boulatar M, Belaid WA, Ahbala T, et al. (2025) Ruptured Hepatic Hydatid Cyst into the Bile Ducts: Case Series Study. J Surg 10:11518 DOI: 10.29011/2575-9760.011518

Received Date: 12 December 2025; **Accepted Date:** 18 December 2025; **Published Date:** 20 December 2025

Abstract

Introduction: Intrabiliary rupture of hepatic hydatid cysts represents the most frequent and serious complication of hepatic echinococcosis. It commonly presents with cholangitis or obstructive jaundice, and diagnosis relies on an integrated clinical, biological, and radiological assessment. The aim of this study was to evaluate the clinical characteristics, the comparative contribution of imaging modalities, and the therapeutic outcomes of this complication.

Materials and Methods: This retrospective study included 50 cases of hepatic hydatid cysts ruptured into the bile ducts, collected over a 4-year period. Epidemiological, clinical, biological, radiological, perioperative, and follow-up data were analyzed. Diagnostic modalities, surgical techniques, and complications were compared with the literature.

Results: Intrabiliary rupture accounted for 25% of operated cysts (50/200). The mean age was 38.5 years with a female predominance (54%). Cholangitis was the main presenting symptom (56%). Ultrasound, performed in 100% of patients, identified the cyst but confirmed the fistula in only 26%. CT scan showed direct or indirect signs in 20%, while MR cholangiography (bili-MRI), performed in 10 patients, accurately mapped the biliary communications. All patients underwent surgery: 90% received conservative treatment and 10% underwent radical surgery. The overall postoperative complication rate was 24%, dominated by external biliary fistulas (14%). No deaths were recorded, and only one recurrence was noted.

Conclusion: Intrabiliary rupture of hepatic hydatid cysts remains a manageable medico-surgical emergency with an appropriate diagnostic and therapeutic strategy. Ultrasound and CT are useful initial examinations, but bili-MRI appears to be the most effective tool for preoperative assessment of the fistula. Conservative techniques yield good outcomes, and integration of Endoscopic Management (ERCP) could improve care and reduce postoperative morbidity. A standardized approach combining bili-MRI and ERCP seems to be the optimal strategy for future management.

Keywords: Biliary Tract Diseases; Biliary Fistula; Choledocholithiasis / Hydatid Debris Migration; Cholangiography; Endoscopic Retrograde Cholangiopancreatography (ERCP); Hydatid Cyst, Hepatic Echinococcosis; Hepatectomy / Conservative Surgical Management; Laparoscopy; Postoperative Complications

Introduction

The hepatic hydatid cyst represents the most frequent localization of human echinococcosis in endemic countries, particularly in the Maghreb, where the persistence of the dog–sheep parasitic cycle contributes to a high infectious reservoir [1]. In Moroccan hospital series, this condition accounts for a significant proportion of hepatobiliary surgical indications, with a non-negligible rate of evolving complications [2-5]. Among these, cysto-biliary rupture is considered the most common complicated form and remains associated with significant morbidity, potentially worsened by infection, cholangitis, or common bile duct obstruction [2-6]. The diagnosis of a biliocystic fistula relies on a combination of clinical, biological, and especially radiological findings. Ultrasound and CT scan serve as first-line examinations, allowing assessment of the cyst's evolutionary stage and identification of indirect signs of biliary disruption [3-6]. However, MRI, particularly the MRCP sequence, provides direct visualization of cysto-biliary communications and intraductal debris, with greater sensitivity for detecting small or occult leaks [7]. This radiological advancement has optimized preoperative fistula detection, guided surgical strategies, and reduced the risk of therapeutic failure [7,8]. Therapeutically, surgery remains the historical standard of care. Commonly used techniques include DITFO, the Perdomo method, external biliary drainage via a T-tube (Kehr), and eradication of hydatid content [8,9]. While effective, these procedures carry persistent risks of residual fistulas, infection, or the need for reoperation [8-10]. Concurrently, the rise of interventional endoscopy has transformed modern decision-making algorithms. ERCP allows extraction of membranes, relief of bile duct obstruction, reduction of biliary leaks, and in some cases, avoidance of immediate surgery [11]. Several published clinical reports have documented successful endoscopic management of ruptured hydatid cysts with biliary fistulas, particularly when the bile duct is patent or the fistula is small [11,12]. A unique Moroccan experience also reports the use of ERCP as a complement or alternative to surgery, demonstrating the effectiveness of a combined approach and reduced postoperative complications in selected cases [13]. Thus, the collective data from international and Moroccan series suggest that the optimal management of ruptured hepatic hydatid cysts into the biliary tract relies on an integrated strategy combining advanced diagnostic imaging, targeted surgical intervention, and interventional endoscopy. The aim of this study is to provide a structured analysis of the diagnostic, therapeutic, and outcome modalities of this complication, based on a cross-analysis of clinical findings and consolidated bibliographic data.

Materials & Methods

This is a retrospective, single-center clinical study conducted in the General Surgery Department of CHU Mohammed VI in Marrakech. It included all patients treated for hepatic hydatid cysts complicated by confirmed intrabiliary rupture over a continuous 4-year period. Data were collected from hospitalization records, operative registers, ERCP reports, and pre- and postoperative imaging studies [1].

Data Collected

Domain	Variables
clinical	Jaundice, right hypochondrial pain, fever, cholangitis
biological	Total/direct bilirubin, ALAT/ASAT, CRP, leukocyte count
imaging	Cyst size, WHO classification, segmental location, bile duct dilation, intraductal membranes
per-operative	Type of rupture (minor vs major), hydatid content, active biliary leak, procedure performed
treatment	Surgery alone / ERCP alone / sequential combined approach
outcome	Persistent fistula, length of hospital stay, complications, reintervention, mortality

Table 1: Studied Domains and Variables

Decision-Making Approach and Analysis

Patients were stratified according to the primary therapeutic approach implemented:

- Primary surgery**;**
- Primary ERCP**;**
- Sequential or combined strategy**.**

This stratification allowed for the assessment of the impact of the initial treatment choice on postoperative morbidity, length of hospital stay, and the need for reintervention. The results were interpreted in the context of international literature [9-12], taking into account the Moroccan endemic setting and the available technical resources. The central methodological objective is to determine under which conditions surgery remains the reference

treatment, when ERCP can serve as an alternative, and how to establish a reproducible therapeutic algorithm in visceral surgery centers managing cysto-biliary fistulas [11-13].

Results

Epidemiological Data

Between May 2016 and May 2020, 200 patients underwent surgery for hepatic hydatid cysts. Among them, 50 presented with rupture into the biliary tract, representing 25% of the operated hepatic hydatid cysts.The mean age of the patients was 38.5 years, ranging from 15 to 80 years. The most affected age group was 20–49 years, accounting for 29 cases (58% of the series).There was a slight female predominance: 54% of patients were female and 46% male, with a male-to-female sex ratio of 0.85. The majority of patients (56%) came from rural areas, compared to 44% from urban areas. Contact with dogs was reported in 36 patients (72% of the series), predominantly among rural patients (27 cases, 75% of contacts) versus urban patients (9 cases, 25%). Fourteen patients (28%) reported no known contact with dogs. Regarding medical history, 5 patients (10%) had a previous hepatic hydatid cyst, and 2 patients (4%) had a history of pulmonary hydatid cyst. In total, 7 patients (14%) had personal histories of hepatic or pulmonary hydatid disease.

Clinical Presentation

The mean duration of symptoms before specialized consultation was 15 days, with extremes ranging from 1 day to 1 month. Symptom onset was gradual in 22 cases (44%) and abrupt in 19 cases (38%), while it was not specified in 9 patients. Abdominal pain was the most frequent reason for consultation. It was localized in the right hypochondrium or epigastrium, and in some cases presented as generalized abdominal pain, reported in 9 patients (19.14%). A picture of acute cholangitis, combining right hypochondrial pain, fever, and obstructive jaundice, was observed in 28 patients (56% of the series). Isolated febrile syndrome (fever and chills) was noted in 8 patients (16%). Cholestatic-type jaundice was frequently observed, either isolated or associated with other symptoms depending on the case. On clinical examination, tenderness of the right hypochondrium was found in 33 patients, generalized abdominal tenderness in 9 patients (19.14%), and guarding of the right hypochondrium and/or epigastrium in 18 patients (36%). Hepatomegaly was present in 15 patients (30%). A palpable, rounded, firm, well-defined mass in the right hypochondrium was described in several cases. No patient presented with splenomegaly.

Biological Data

Hydatid serology was performed when available and was positive in 23 cases, representing **76.66%** of patients.

The complete blood count revealed:

- **Leukocytosis** in 36 patients (72%), most often reflecting cyst infection or rupture into the biliary tract;
- **Eosinophilia** in 6 patients (12%);
- **Combined leukocytosis and eosinophilia** in 3 patients (6%);
- **Anemia** in 5 patients (10%).

An elevated **C-reactive protein (CRP)** and a biological inflammatory syndrome were frequently associated with acute cholangitis.

Imaging

Ultrasound

Abdominal ultrasound was performed in all patients. It enabled the diagnosis of hepatic hydatid cysts and allowed precise determination of their location and echographic type in every case.

Regarding the number of cysts:

- A single hepatic hydatid cyst was observed in 30 patients (60%).
- Two cysts were found in 12 patients (24%).
- Multicystic forms (three or more cysts) were less common, including one patient (2%) with multiple hydatid cysts.

The ultrasound appearances were classified according to **Gharbi's classification**; several patients had mixed types or a combination of two or more types, noted in 6 cases (12%).

Ultrasound also assessed the biliary tract and allowed:

- Suspecting cysto-biliary rupture in 30 cases, indicated by the association of a hepatic hydatid cyst and dilated bile ducts.
- Confirming the diagnosis in 13 cases by simultaneously showing biliary dilatation and the presence of intrabiliary hydatid material.

Computed Tomography (CT)

Abdominal CT was performed in all patients. It confirmed the diagnosis of hepatic hydatid cyst (HHC) in 27 cases previously suspected on ultrasound. In 3 cases, CT did not detect biliary abnormalities that had been seen on ultrasound. In 7 cases, CT only showed the hepatic cysts without associated biliary anomalies. Overall, CT enabled the diagnosis of cysto-biliary rupture in 10 cases (20%) by identifying either direct or indirect signs of a fistula. Among these signs, intrahepatic bile duct and common bile duct dilation was consistently observed. Visualization of hydatid material in the common bile duct was noted in some cases, while

in others it was only detected intraoperatively. One case presented with acute pancreatitis, Balthazar stage B.

Magnetic Resonance Cholangiopancreatography (MRCP)

MRCP was performed in 10 patients (20% of the series) who had signs of cholangitis and significant biliary dilatation. This imaging was used to better delineate the relationships between the hydatid cyst, the biliary tract, and the vascular pedicle, and to detect cysto-biliary fistulas. MRCP allowed detailed visualization of biliary communications and aided therapeutic planning in these cases.

Surgical Treatment

All patients in this series underwent surgical management. Those admitted with acute cholangitis or pancreatitis first received preoperative resuscitation, including fluid and electrolyte rebalancing, antibiotics, correction of coagulation disorders, and analgesics. In these patients, the timing of surgery ranged from day 3 to day 14 of hospitalization, with an average of 8 days. For other patients, the intervention occurred earlier, between day 1 and day 8, with a mean of 4 days.

The surgical approaches used were:

- Right subcostal incision in 38 cases (76%)
- Upper midline incision in 8 cases (16%)
- Right subcostal incision extended to the left in 3 cases (6%)
- Bilateral subcostal incision in 1 case (2%)

Conservative surgical techniques were employed in the vast majority of cases, primarily consisting of partial cystectomy (resection of the protruding dome). Radical procedures were performed in 5 patients, including 3 total pericystectomies and 2 left hepatectomies, addressing both the hydatid cyst and the biliary fistula.

Management of Biliary Fistula

The biliary fistula was most often managed conservatively. The techniques employed included:

- Direct suture (closure) of the fistula in 24 cases (48%)
- Catheterization of the fistula in 2 cases (4%)
- Transhepatic cysto-biliary drainage according to Perdomo in 5 cases (10%)
- Biliary-digestive anastomosis in 1 case (2%), specifically a choledochoduodenal bypass
- Unipolar drainage in 4 cases (8%)
- Bipolar drainage in 3 cases (6%)
- Common bile duct drainage using a Kehr tube combined with fistula closure in 10 cases (20%)
- Fistula left untreated due to small caliber in 1 case (2%)

The residual cavity was systematically drained via subhepatic and/or inter-hepato-diaphragmatic drains. Additional techniques for residual cavity management included epiplooplasty in 1 case and capitonnage in 6 cases.

Cholecystectomy was performed in 20 patients (40%), 10 for associated gallstones and 10 for gallbladder fistula.

Morbidity, Mortality, and Outcomes

No deaths were reported in this series.

Postoperative recovery was uneventful in 35 patients. Postoperative complications occurred in 12 patients (24%), including:

- External biliary fistula in 7 patients (14%)
- Surgical site infection in 1 patient (2%)
- Isolated fever in 3 patients (6%)
- Peritonitis in 1 patient (2%)

The mean hospital stay was 10 days, ranging from 5 to 30 days. Postoperative hospitalization after surgery ranged from 3 to 14 days depending on recovery and drainage requirements.

During long-term follow-up, recurrence of hepatic hydatid cyst was observed in only one patient.

Discussion

Rupture of Hepatic Hydatid Cysts (HHC) into the biliary tract represents the most frequent and severe complication of hepatic echinococcosis. Our study found an incidence of 25% among operated HHCs, which aligns with data from large published series reporting rates ranging from 13% to 42%, depending on the cohort and the level of surgical selection [1,2]. This variability can be explained by differences in population characteristics, parasitic exposure between rural and urban areas, and the heterogeneity of diagnostic criteria used in the literature.

Comparative Epidemiological and Clinical Profile

The mean age observed in our series (38.5 years) aligns with trends reported in several studies, which note a predominance between 30 and 50 years [3,4]. The slight female predominance we observed is also reported in other North African series, likely related to the distribution of livestock-related activities and exposure to dogs [5]. The high frequency of acute cholangitis (56%) underscores the severe and overt nature of this complication and is consistent with

the rates reported by Petrakis et al., who noted a septicobiliary presentation in more than half of cases with biliary fistula [6,7]. The high prevalence of dog contact (72%) further emphasizes the zoonotic and preventive aspects of the condition.

Diagnosis and Comparative Contribution of Imaging Studies

Ultrasound enabled the diagnosis of hepatic hydatid cysts in all patients, but confirmed the cysto-biliary fistula in only 26% of cases. This diagnostic limitation is consistent with the literature, as ultrasound is effective for detecting the cyst itself but has low sensitivity for direct visualization of biliary communications [8,9]. CT showed direct signs of biliary leakage in 20% of cases, reflecting intermediate sensitivity in our series. This aligns with international data, where CT detects cysto-biliary fistulas in 25–50% of cases depending on the cohort [10]. Biliary MRI (MRCP), performed in 10 patients, proved decisive for preoperative mapping of cysto-biliary communications. Several recent studies highlight MRCP as the reference imaging modality when available [11,12]. Overall, our findings confirm that while ultrasound and CT serve as useful initial diagnostic tools, MRCP remains the most accurate modality for precise characterization of the fistula, directly guiding therapeutic strategy.

Therapeutic Approach: Surgery as the Mainstay, Yet Open for Discussion

In our cohort, surgery was performed in 100% of patients—reflecting the local context but contrasting with international trends where ERCP increasingly plays a pre- or post-operative role. Conservative techniques predominated (90%), reflecting the aim to preserve hepatic parenchyma and limit aggressive interventions. Clinical outcomes align with the literature, where surgical series report fistula control in 80–95% of cases, albeit with a notable risk of residual biliary morbidity. Radical procedures (total pericystectomy, left hepatectomy, n = 5) were reserved for complex cases, consistent with international recommendations that advocate radical treatment only in the presence of multiple, infected, or recurrent hydatid cysts.

Postoperative Morbidity and Critical Analysis

The postoperative complication rate (24%), including 14% persistent fistulas, aligns closely with the literature, where residual external fistula rates range from 10 to 30% depending on the procedure performed and the inflammatory status of the biliary pedicle. The absence of mortality in our series is a strength but should be interpreted cautiously given the sample size and the lack of comprehensive long-term follow-up.

Study Limitations and Practical Implications

This study is single-center, retrospective, and based on hospital records, limiting its generalizability. The lack of systematic access to ERCP also prevents direct comparison with more modern strategies. However, the cohort size (N = 50) and standardized surgical approach represent major methodological strengths.

Immediate Clinical Implication

A decision-making protocol incorporating bili-MRI (+/- ERCP depending on availability) could reduce postoperative fistulas by better identifying preoperative biliary communications.

Future Perspective

A prospective study comparing surgery alone versus surgery combined with ERCP and optimized bili-MRI represents a logical next step following this work.

Conclusion

Cysto-biliary rupture of hepatic hydatid cysts represents a frequent, severe complication with high morbidity potential. Our series of 50 cases confirms that acute cholangitis is the predominant clinical presentation, reflecting acute biliary obstruction by hydatid material. Ultrasound remains a systematic first-line examination but has diagnostic limitations, whereas bili-MRI appears to be the most effective tool for detecting fistulas and guiding therapeutic decisions. Treatment remains primarily surgical in our setting, favoring conservative techniques that yield satisfactory results, albeit with a notable rate of residual external fistulas. The absence of mortality and the low recurrence rate confirm the effectiveness of the employed strategies. Greater systematic integration of bili-MRI and interventional endoscopy (ERCP) could further reduce morbidity and optimize management. Although retrospective and single-center, our study provides a clear contribution to structuring the diagnosis and treatment of cysto-biliary fistulas and paves the way for prospective comparative protocols systematically incorporating advanced imaging and ERCP.

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