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# Evaluation of Imaging Findings in Hepatic Hydatid Cyst Patients with Intrabiliary Rupture: A Retrospective Magnetic Resonance Imaging Study

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## Abstract

**Background:** Liver hydatid cyst ruptures entering the biliary tree can lead to life-threatening conditions. This study investigates the frequency of biliary rupture and the most common magnetic resonance imaging findings.

**Methods:** Patients with hydatid cyst within the last 5 years were screened. Demographic data, the hepatic segment location, number of lesions, lesion size, and type of cyst were recorded, and biliary communication was evaluated. In patients with biliary rupture, the presence of dilation bile ducts, the presence of structures consistent with cyst contents in the bile ducts, the presence of fistulous connections, and the frequency rates of these findings were evaluated.

**Results:** Fifty-five cyst hydatid lesions from 50 patients (30 male and 20 female) were included. 4 (8%) patients had biliary rupture. Among them 4 (100%) patients had dilation in the intrahepatic and common bile ducts, 3 (75%) had deformation of cyst, 2 (50%) had structures consistent with cyst contents in the even ducts, and 1 (25%) had a fistulous connection shown by magnetic resonance imaging. Age and gender did not differ between patients with and without biliary rupture ($P > .05$). Patients with biliary rupture had larger cysts than those without ($P < .05$), and type 3 hydatid cysts were the most common.

**Conclusion:** In conclusion, the frequency of intrabiliary rupture in our study was found to be 8%, and significant imaging findings can be obtained with magnetic resonance imaging in the evaluation of intrabiliary rupture. Bile duct dilation, cyst deformation, cyst contents in the bile ducts, and fistulous connection are the most common magnetic resonance imaging findings in patients with intrabiliary rupture.

**Keywords:** Cyst hydatid, Echinococcosis granulosus, rupture, cystobiliary fistula, cystobiliary communication, intrabiliary rupture, magnetic resonance imaging

## INTRODUCTION

Hydatid cysts are parasitic infections caused by *Echinococcus* larvae. Hydatid cyst disease affects various regions of the world. There are 2 primary types of hydatid cysts, namely *Echinococcus granulosus* and *Echinococcus multilocularis*. Humans are fortuitous intermediary hosts and typically contract the illness through indirect exposure. The cyst can grow in any organ of the body,[1] but it is most commonly found in the liver (60%-70%) and lungs (20%-30%). The infection is acquired by the ingestion of tapeworm eggs, which are shed in the feces of canines and other carnivores. The eggs undergo hatching inside the limits of the small intestine and subsequently migrate to other organs in the body. Upon reaching their ultimate location, the larvae form cysts.[2-5]

Patients with symptomatic hydatid liver cysts typically present with upper abdominal pain and a loss of appetite; bile duct compression may result in jaundice. Palpation may reveal a tumor-like mass, hepatomegaly, or distension in the abdomen. Cysts in the lung can cause pain in the chest, coughing, or hemoptysis, and cyst rupture into the bronchi can result in the evacuation of hydatid materials. Cyst rupture in any organ can cause fever, urticaria, eosinophilia, and anaphylactic shock. Any puncture of a hydatid cyst lesion is contraindicated due to the possibility of potentially lethal allergic reactions resulting from cyst rupture.[5,6]

Cyst hydatid is diagnosed based on clinical signs, imaging studies, and serology tests. The utilization of routine imaging or population-based ultrasound (US) screening of the liver has the potential to detect asymptomatic cysts, making it a crucial practice for the early identification of patients in regions with a high prevalence of this condition.[6-8] Imaging modalities play a crucial role in the diagnostic process, wherein the cost-effective and easily transportable US is commonly employed for the detection of cyst hydatid liver lesions. On the other hand, x-ray imaging is utilized for the diagnosis of lung cysts. Both procedures are employed for the purposes of diagnosis, population screening, and subsequent monitoring. Abdominal US, computed tomography (CT), and magnetic resonance imaging (MRI) are employed for the purpose of diagnosing complications. Recently, the use of dual-energy CT has been reported.[9] In contemporary medical practice, the utilization of endoscopic retrograde cholangiopancreatography (ERCP) has become widespread for both the identification and the management of biliary complications associated

**Table 1.** Magnetic Resonance Imaging Sequence Parameters

| | Non-Contrast Scans | | | | | Contrast-Enhanced Scans |
|---|---|---|---|---|---|---|
| **MRI Sequences Type of Scan** | **Ax T2 SSFSE BH** | **Ax T2 SSFSE ASPIR BH** | **Cor T2 SSFSE BH** | **Ax Dual Echo BH** | **Ax DWI b0-b800 Rtr** | **+C Ax LAVA Dyn BH** |
| TR (ms) | 1000 | 1000 | 1200 | 200 | 4500<br>6000 | 6.4 |
| TE (ms) | 85 | 85 | 85 | 2.3<br>4.6 | 80<br>120 | 2.1 |
| NEX | 1 | 1 | 1 | 1 | 1 | 1 |
| FOV ($mm^2$) | 340 × 340 | 340 × 340 | 360 × 360 | 320 × 320 | 340 × 340 | 340 × 340 |
| Slice thickness (mm) | 6 | 6 | 6 | 4 | 6 | 4 |

Ax, axial; Cor, coronal; DWI, diffusion weighted imaging; FOV, field of view; MRI, magnetic resonance imaging; TE, echo time; TR, repetition time; T2, T2-weighted image.

with hepatic hydatid cysts.[3] The diagnostic value of MRI looks to be better compared to that of CT scan in cases of cyst hydatid. Both imaging techniques, however, are complementary in the diagnosis of cyst hydatid and should be employed in order to obtain adequate information for making therapeutic decisions. Nevertheless, it is worth noting that T2-weighted MRI microcystic images are considered to be pathognomonic indicators of cyst hydatid lesions.[10-13]

Although there is a link between the biliary system and the cyst in around 80%-90% of individuals with hepatic hydatid cysts, the occurrence of clinical biliary rupture is rather low, ranging from 13% to 37%. Due to the high mortality and morbidity of biliary rupture, its management becomes crucial. However, the diagnosis of occult intrabiliary rupture, which accounts for 10%-37% of cases, is challenging due to the lack of notable symptoms and preoperative radiological findings.[4] Biliary rupture represents the prevailing complication associated with hepatic hydatid cysts, manifesting in around 14%-25% of individuals who experience postoperative bile leakage.[14] The most prevalent complications associated with anaphylactic shock include cyst infection of the biliary tract and rupture into the peritoneal space. The occurrence of intrabiliary rupture has been documented to range from 6.1% to 17%.[4,15] In a research conducted during a period when the US and CT were not accessible, the reported rate was found to be 41%.[16]

The aim of this study was to investigate the frequency of intrabiliary rupture in patients with hepatic hydatid cysts and evaluate the most common MRI findings in patients with biliary rupture.

## METHODS

Ethical approval was obtained from the Kastamonu University Hospital's Ethics Committee for this retrospective study (Date: July 5, 2023, Number: 2023-KAEK-78).

### MAIN POINTS

- Due to the high mortality and morbidity of biliary rupture, its management becomes crucial.
- The frequency of cystobiliary fistula in our study was found to be approximately 8%.
- The most commonly observed magnetic resonance imaging finding in cystobiliary fistula is bile duct dilation, along with other important findings such as cyst deformation, structures consistent with cyst contents in the bile ducts, and fistulous connection.

The reports of patients who underwent upper abdominal and liver MRI within the last 5 years were screened using the hospital's information management system with the keyword "hydatid cyst."

The inclusion criteria were defined as follows: positive diagnosis of hepatic hydatid cyst, images obtained using a protocol specific to the liver, images free of artifacts suitable for evaluation, and being 18 years of age or older.

The exclusion criteria were defined as follows: being under 18 years of age, having additional liver pathologies that could affect the study results such as diffuse liver parenchymal disease or malignancy, having undergone surgical intervention involving the gallbladder or bile ducts, images with artifacts, MRI performed without the use of contrast agent, and images obtained without an appropriate protocol specific to the liver.

All examinations were performed using a 1.5 Tesla MR device (Signa Explorer, GE Medical Systems, Milwaukee, Wis, USA). The sequence parameters are shown in Table 1.

In addition to demographic data such as age and gender of the scanned patients, information regarding the hepatic segment location of the hydatid cyst lesion, number of lesions, lesion size, and type of cyst according to the Gharbi classification was recorded. Furthermore, the presence or absence of biliary communication was evaluated, and the clinical and laboratory findings of these patients were checked from the hospital information system.

In patients with biliary communication, the presence of dilation in the intrahepatic bile ducts and common bile duct, the presence of structures consistent with cyst contents in the bile ducts, the presence of fistulous connections, and the frequency rates of these findings were evaluated. Additionally, the ERCP findings of patients with biliary communication were retrospectively evaluated from the hospital information management system.

### Statistical Analysis

The statistical program used for all analyses was IBM Statistical Package for the Social Sciences 26.0 (IBM SPSS Corp., Armonk, New York, USA). Descriptive statistics are presented as mean ± SD for numerical data as well as counts and percentages for categorical data. The normality of the data was evaluated through the utilization of the Kolmogorov–Smirnov test, which revealed that none of the variables exhibited adherence to a normal distribution. Therefore, in order to compare groups, nonparametric tests such as the Mann–Whitney *U*- test are employed. A *P*-value <.05 was considered to be statistically significant.

## RESULTS

As a result of the screening conducted through the hospital information system, the study included a total of 50 patients who met the predetermined criteria for inclusion.

Among the 50 patients, 3 of them had 2 lesions each and 1 patient had 3 lesions. In total, there were 55 lesions. The mean age of the patients was 44.7 ± 10.4 years. Of the 50 patients, 30 (60%) were male and 20 (40%) were female. Age and gender values are shown in Table 2.

**Table 2.** Patients' Gender, Age, and Lesion Number Values and Comparison of These Values by Gender

| | Male (n = 30) | Female (n = 20) | Total (n = 50) | *P* |
|---|---|---|---|---|
| Age (years) | 46.7 ± 11.3 | 42.9 ± 9.1 | 44.7 ± 10.4 | .547 |
| Number of lesions | 31 | 24 | 55 | .354 |

Thirty-two (58.2%) of 55 hydatid cyst lesions were located in the right lobe and 23 (41.8%) were located in the left lobe. The mean lesion diameter was 45.8 ± 29.6 mm. There was no significant difference between the lesions in the right and left lobes in terms of lesion sizes

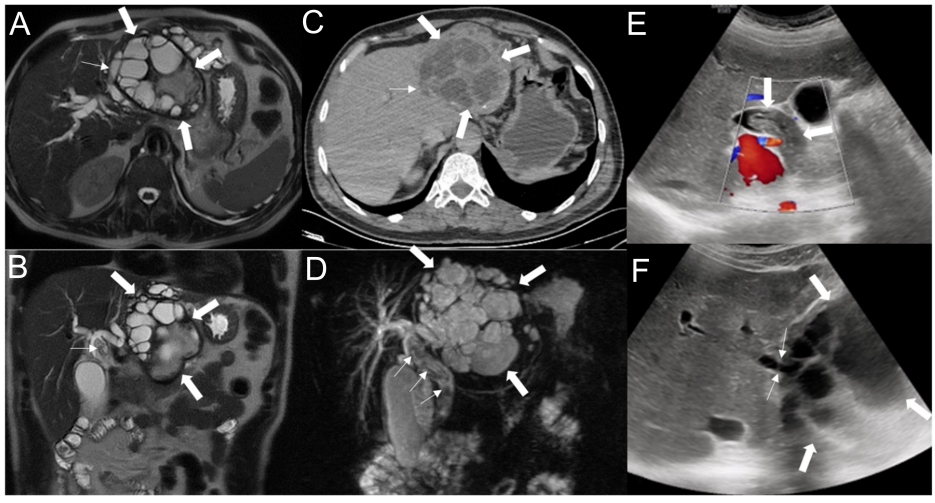


**Figure 1.** Fifty-four-year-old male with Gharbi type 3 hydatid cyst lesion in the left lobe of the liver with 6-day epigastric stomach pain and dyspeptic symptoms. (A, B) Axial/coronal T2-weighted fast spin echo magnetic resonance images demonstrate a type 3 hydatid cyst lesion (thick arrows). The common and main hepatic bile ducts contain hypointense membranous structures connected to hydatid cysts and are dilated. (C) Axial abdominal contrast-enhanced computed tomography scan shows a type 3 hydatid cyst (thick arrows) and a fistulous connection between it and one of the intrahepatic bile ducts (thin arrow). (D) Coronal magnetic resonance cholangiopancreatography image shows a type 3 hydatid cyst lesion (thick arrows). There is also bile duct dilatation and hypointense membranous structures caused by hydatid cysts in the common and main hepatic bile ducts (thin arrows). (E, F) Grayscale ultrasound (US) and color Doppler US images show semi-solid structures with uneven heterogeneous supply and no blood supply in the main hepatic bile duct in (thick arrows).

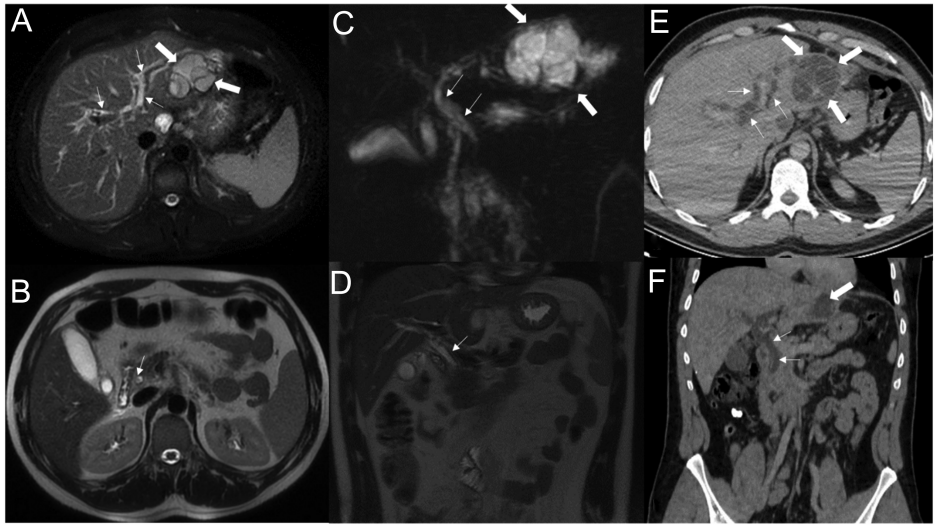


**Figure 2.** Twenty-six-year-old male patient with epigastric abdominal pain that had been ongoing for 5 days and had a Gharbi type 3 hydatid cyst lesion in liver segment 2**.** (A, B) Axial T2-weighted fast spin-echo magnetic resonance images show a hydatid cyst lesion (thick arrows) and dilatation of the bile ducts (thin arrows). (C, D) Coronal magnetic resonance cholangiopancreatography images and coronal T2-weighted fast spin-echo magnetic resonance images show a hypointense membranous structure related to the hydatid cyst in the lumen of the common bile duct (thin arrows) and show hydatid cyst lesion (thick arrows). **(**E, F**)** The axial and coronal abdominal contrast-enhanced computed tomography images of the patient show a hydatid cyst lesion (thick arrows) and dilatation of the bile ducts (thin arrows).

**Table 3.** Magnetic Resonance Imaging Findings in Patients with Intrabiliary Rupture

| Findings | N | % |
|---|---|---|
| Dilation in the intrahepatic bile ducts and common bile duct | 4 | 100 |
| Deformation of cyst | 3 | 75 |
| Structures consistent with cyst contents in the bile ducts | 2 | 50 |
| A fistulous connection | 1 | 25 |

($P > .05$). Out of the 55 lesions, 10 (18.2%) were classified as type 1 according to the Gharbi classification, 4 (7.3%) were type 2, 6 (10.9%) were type 3, 15 (27.3%) were type 4, and 20 (36.4%) were type 5, consistent with hydatid cysts.

Out of the 50 patients, 4 (8%) had biliary communication. In addition to the MRI findings of all patients, the ERCP results were also consistent with biliary communication. All patients with biliary communication had a single lesion. Among the 4 patients with biliary communication, 4 (100%) had dilation in the intrahepatic bile ducts and common bile duct, 3 (75%) had deformation of cyst, 2 (50%) had structures consistent with cyst contents in the bile ducts, and 1 (25%) had a fistulous connection shown by MRI (Figures 1 and 2). The data of patients with biliary communication are shown in Table 3.

No significant differences were found in terms of age and gender between patients with biliary communication and those without ($P > .05$). However, the cyst size was significantly larger in patients with biliary communication compared to those without ($P < .05$). In terms of the cyst type, the most common type of hydatid cyst in patients with biliary communication was type 3.

## DISCUSSION

In our study, we retrospectively evaluated the frequency of intrabiliary rupture in patients with hydatid cyst, the factors influencing rupture, and the most common MRI findings in cases with rupture. According to our study, the most common MRI finding in hydatid cyst with biliary communication is biliary dilation. Following that, the presence of cystic material within the bile ducts and direct demonstration of fistulous connections are observed in descending order of frequency.

Rupture of cysts into the bile ducts is a common complication of hepatic hydatid disease. Communication between the biliary tree and the hydatid cyst can be either evident or occult. If an occult cystobiliary communication remains undetected and is not properly repaired during surgery, it can result in postoperative biliary fistulas. Imaging plays a crucial role in the detection of hydatid cysts and accurate localization of intrabiliary ruptures, which is essential for early surgical intervention.[16,17]

Prior to the utilization of US and CT, preoperative identification of hepatic hydatid cyst complications was challenging and relied on clinical symptoms and laboratory results. Ultrasound or CT can often indicate the presence of overt intrabiliary rupture, while MRI offers the advantage of additional multiplanar images. Magnetic resonance cholangiopancreatography (MRCP) reveals distinct features such as a daughter cyst, dilated biliary tree containing hydatid materials, prominent border, and membrane separation.[18,19]

Rupture of hydatid cysts can occur due to increased pressure inside the cyst triggered by trauma, exertion, or forceful coughing. The rupture can be categorized into 3 types: contained rupture, communicating rupture, and direct rupture. In a contained rupture, the endocyst ruptures, but the contents of the cyst are still confined within the pericyst. Communicating rupture involves the rupture of the endocyst, resulting in the leakage of cyst contents through smaller biliary ducts. Direct rupture occurs when the 2 layers of endocyst and pericyst rupture, causing the cyst contents to leak into the peritoneal or pleural spaces.[3,4]

The radiological findings of intrabiliary rupture encompass both direct and indirect signs.[20] The sole direct sign of rupture is evident communication between the cyst and the biliary tree. In our study, this finding was positive in only 25% of cases with ruptured cysts. Similarly, Erden et al[17] reported a relatively low rate of 33% in their study, which assessed MRCP for cystobiliary communication in a population of 54 hydatid cyst patients and 12 cystobiliary communication patients. Although the presence of this finding indicates a high probability of cystobiliary rupture, it can be relatively low in detection by MRI and MRCP, as demonstrated in our study and literature.

The cyst deformation, which is one of the indirect signs of intrabiliary rupture and considered as a result of decreased intracystic pressure, was found positive in 75% of cases with cystobiliary communication in our study. Our findings are consistent with the study conducted by Erden et al,[17] which reported the same rate of 75%. This finding may also be secondary to decreased cyst pressure in patients who have undergone treatment.[21] However, none of the patients in our study had a history of treatment. Although this sign does not definitively indicate rupture, it can be considered as a highly indicative indirect finding of cystobiliary connection based on our study and the literature. The identification of collapse or deformation in a known hydatid cyst during follow-up and comparison with previous images can be considered as a significant indirect finding of cystobiliary fistula.

The pericyst layer, consisting of thick avascular and fibrous tissue, is located on the outermost part of the hydatid cyst lesion.[22] Loss of integrity in this pericyst layer and demonstration of communication with the bile ducts are direct signs of biliary rupture. Discontinuity in the pericyst layer was relatively low in our cases and found in only about 25% of cases. In the literature, it has been reported that this finding was present in approximately 66.7% of cases in the study conducted by Erden et al.[17] The relatively lower percentage in our study could be attributed to a lower number of positive cases for cystobiliary fistula. Nevertheless, the detection of defects in the pericyst, especially in T2-weighted series, and loss of integrity in the cyst wall should be emphasized as significant findings of rupture.

It has been stated that the inner layer of a hydatid cyst, known as the endocyst, can detach not only due to aging or ischemia but also in cases of rupture.[23] In the study by Erden et al,[17] it was reported that endocyst detachment was observed in 50% of cases. However, we did not observe this finding in any of our cases. Additionally, the presence of cyst fragments within the bile ducts due to biliary communication is another finding, and we observed this in 50% of our cases. In the study by Erden et al,[17] they reported a 25% incidence of this finding. Variations in the visibility of cystic contents in the bile ducts on MRI may arise depending on the size and location of the cyst and the degree of rupture.

In our study, all cases with intrabiliary rupture showed dilation in the intrahepatic bile ducts and common bile duct. In the literature, Wani et al[24] reported findings from a study evaluating CT findings in 6 patients

with intrabiliary rupture, stating that, similar to our study, all patients showed dilation in the bile ducts. Additionally, dilation in the intrahepatic bile ducts can sometimes be observed in pericystic areas due to an increase in the cyst size. Focal dilation and localized pericystic areas can sometimes be falsely interpreted as intrabiliary rupture, as reported in the literature.[17] However, in our study, the dilation of the bile ducts in the cases was not localized but rather generalized, and the intrabiliary rupture was confirmed by ERCP without any false-positive results.

Our study had some limitations. The most important limitation was its retrospective design and the relatively small population of patients with hepatic hydatid cysts who underwent abdominal MRI. Additionally, the relatively low number of cases with cystobiliary fistula was also a significant limitation of the study. Furthermore, due to the retrospective design, sufficient data regarding the clinical symptoms of the patients could not be obtained, which was another limitation of the study. However, a strong aspect of the study was that all 4 cases with cystobiliary fistula were confirmed by ERCP.

In conclusion, the frequency of cystobiliary fistula in our study was found to be approximately 8%, and significant imaging findings can be obtained with MRI in the evaluation of cystobiliary fistula. The most commonly observed MRI finding in cystobiliary fistula is bile duct dilation, along with other important findings such as cyst deformation, structures consistent with cyst contents in the bile ducts, and fistulous connection. Longitudinal prospective studies with a larger patient population could provide additional benefits in this regard.

**Ethics Committee Approval:** Ethics committee approval was received for this study from the ethics committee of Kastamonu University (Date: July 5, 2023, Number: 2023-KAEK-78).

**Informed Consent:** Due to the retrospective design of the study, patient consent was not obtained for this study.

**Peer-review:** Externally peer-reviewed.

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