

# Complications of Hydatid Cyst of Liver and its Management: A Review

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## ABSTRACT

Hydatid disease of the liver is still endemic in certain parts of the world. The diagnosis of non-complicated hydatid cyst of the liver depends on clinical suspicion. The modern treatment of hydatid cyst of the liver varies from surgical intervention to percutaneous drainage or medical therapy. Surgery is still the treatment of choice and can be performed by the conventional or laparoscopic approach. However, hydatid cysts may present with complications such as cysts ruptured into the biliary tract (12%), cysts rupture involving the lung or pleural space (2.2%), cysts ruptured into the peritoneum (1.6%), and cysts ruptured into the digestive tract (0.2%). Acute cholangitis is the most common syndrome, as a result of the presence of the ruptured hydatid cystic contents into the biliary tract. Hence, this review article will focus on the complications of hydatid cyst of liver and its management approach.

**Keywords:** Biliary, Cyst, Liver, Management, Peritoneum

*Asian Pac. J. Health Sci.*, (2021); DOI: 10.21276/apjhs.2021.8.3.19

## INTRODUCTION

Hydatid disease is a parasitic infestation of humans caused by *Echinococcus granulosus* and *Echinococcus multinocularis* and it is endemic usually in raising countries, but also in most developed countries in Mediteranean region, South America, Africa, etc.<sup>[1]</sup> Dogs and some wild carnivores such as foxes are definitive hosts, harboring worms in their intestine.<sup>[2]</sup> Eggs are passed in the feces and eaten by the intermediate hosts, and larvae encyst in the liver, lungs, and other organs.<sup>[1]</sup>

The liver is the most commonly affected organ with an infestation rate of 60–75%. Specifically, the right hepatic lobe is affected in 80% of cases and the left lobe in 20%. Less common sites are the lungs (15%), spleen, peritoneum, kidneys, brain, etc.<sup>[3]</sup> The hydatid cyst is composed of three layers. The outer layer is the pericyst, an avascular layer derived from modified host tissue and inflammatory cells. The middle layer is a laminated acellular membrane and the inner layer is the germinal layer which produces the laminated membrane and the scolices. The two last membranes form the endocyst.<sup>[4]</sup> Hydatid disease is mostly asymptomatic and many hydatid cysts represent incidental clinical or radiological findings.<sup>[5]</sup>

Complications of hepatic hydatid cysts are uncommon but some, in particular, the complications of liver hydatid cysts are mostly attributed to cyst growth and mass effect on surrounding structures, cyst rupture, or secondary infection.<sup>[6]</sup> The most common complication is the intrabiliary rupture of the hydatid cyst.<sup>[7]</sup> Other less common complications are the rupture of the cyst into the peritoneal cavity, rupture into the thoracic cavity through the diaphragm and toward organs of the gastrointestinal tract and the secondary bacterial infection of the cyst.<sup>[8]</sup> Although surgical management is implicated as the treatment of choice for uncomplicated hydatidosis of the liver, evidence is not clear concerning the management of the biliary tract.<sup>[9]</sup>

## DISCUSSION

It is not uncommon to encounter complications in a patient diagnosed with liver hydatid disease. The reported incidence

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**How to cite this article:** Singh K. Complications of Hydatid Cyst of Liver and its Management: A Review. *Asian Pac. J. Health Sci.*, 2021;8(3):114-118.

**Source of support:** Nil

**Conflicts of interest:** None.

**Received:** 20/04/2021 **Revised:** 29/05/2021 **Accepted:** 05/06/2021

varies from one-third to approximately 60% of the total number of patients. Intrabiliary rupture and cyst super infection are considered to be the most common complications.<sup>[2]</sup>

## Complications of Hydatid Cyst of the Liver

Echinococcal cysts of the liver can cause complications in about 40% of cases. The most common complications of frequency are infection, rupture to the biliary tree; rupture to the peritoneal cavity; and rupture to the pleural cavity. However, rupture in the gastrointestinal tract; bladder and the vessels are very rare.<sup>[10]</sup>

## Superinfection

The superinfection probably occurs from sites next to the hydatid cyst (e.g., biliary or bronchial tree) or as a complication of bacteremia of any cause; secondary bacterial infection has been reported in only 5–8% of cases.<sup>[6]</sup> The evolution of an infected hydatid cyst is usually latent, subacute and is clinically translated by pains in the right hypochondrium, hepatomegaly, and fever.<sup>[10]</sup> Bacteremia arising from a variety of sites is another possible origin of hydatid cyst superinfection.<sup>[2]</sup> The incidence of hepatic hydatid cyst superinfection seems to present with a higher frequency, up to 24%, in large case series of surgically managed cases. Computed tomography is considered the modality of choice

for showing infected liver hydatid cysts. Findings suggestive of infection are a poorly defined lesion, a hyper enhancing rim on a contrast-enhanced exam, patchy enhancing areas in the vicinity of the lesion, and internal gas or air-fluid levels within the cyst. The definite diagnosis is based upon the aforementioned imaging findings in conjunction with the clinical and laboratory signs of pain, fever, and leukocytosis in a patient with a known history of liver hydatid cyst.<sup>[11,12]</sup>

### Intrabiliary Rupture of Hydatid Cyst<sup>[9,10,13,14]</sup>

One of the major and potentially fatal complications of liver echinococcal cysts is their rupture into the biliary tree. This is characterized by the presence of hydatid debris into the common bile duct. It may occur in two forms: An occult rupture, in which only the cystic fluid drains to the biliary tree and is observed in 10–37% of the patients; and frank rupture, which has an overt passage of intracystic material to the biliary tract and is observed in 3–17% of the patients. The incidence of major biliary communications ranges from 5% to 10%. Predisposing factors for intrabiliary rupture are considered to be the older age, larger dimensions, multiplicity of the cysts, and bilobar cysts. Rupture into the biliary tract system tends to occur into the right (55–69.5%), or the left hepatic duct (25–30%), whereas a small percentage (10%) concerns the hepatic duct junction, common bile duct, cystic duct, or the gallbladder. The rupture of the hydatid cyst in the biliary ducts and the migration of the hydatid material in the biliary tree lead to the apparition of other biliary complications such as: Cholangitis, sclerosis odditis, and hydatid biliary lithiasis.

When ruptured into biliary tree, hydatid cysts commonly manifest with findings of biliary obstruction and cholangitis. Diagnosis of this complication can usually be made using ultrasound and abdominal CT scan. Dilatation of intra- or extra-hepatic bile ducts, the presence of hydatid sand and debris into the common bile duct can be imaging signs of a biliary communication. Nevertheless, the only direct sign of a cystobiliary communication is a visible cystic wall defect or the demonstration of the exact location of the communication between the cyst and the biliary tree. Endoscopic retrograde cholangiopancreatography (ERCP) has also proved to be a valuable diagnostic tool for the diagnosis of frank rupture into the biliary tree. Its sensitivity ranges from 86% to 100%. ERCP demonstrates the exact place of the rupture and contributes substantially to the surgical planning, and permits evaluation of acute conditions, such as acute cholangitis and obstruction. Magnetic resonance cholangiography nowadays represents a noninvasive diagnostic alternative modality without the risks associated with ERCP. It depicts the communication of an echinococcal cyst with the biliary tract system, as well as the specific details of its structure.

### The Rupture in the Peritoneum<sup>[15-17]</sup>

Rupture of the cyst in the peritoneal cavity is rare and generally followed by anaphylactic reactions. The reported incidence of this complication ranges from 10% to 16%. A vital hydatid cyst tends to grow in the direction of the least resistance, and usually reaches the liver surface before presenting with enormous proportions. The superficial portion of the pericyst is stretched, thinned out, and the cyst becomes visible as an irregularly shaped, fibrous structure protruding from the normal liver parenchyma. Cysts located in the inferior and anterior part of the liver continue to

grow, protruding into the abdominal cavity. Due to the high intracystic pressure, both univesicular and multivesicular cysts can rupture. As a result, brood capsules, protoscoleces, and daughter cysts from the ruptured hepatic echinococcal cyst are released into the peritoneal cavity. The absorption of the quite toxic antigens of the hydatid fluid from the systemic blood circulation inevitably leads to systemic anaphylactic reaction. Abdominal pain, nausea, vomiting, and urticaria are the most common clinical symptoms. During the clinical examination, signs of acute abdomen such as guarding, rebound, and tenderness are generally present. In silent rupture, the patient develops disseminated abdominal hydatidosis. The release of the hydatid fluid throughout the abdominal cavity results in the development of multiple cysts in the peritoneal cavity with intestinal obstruction, gross abdominal distention, and ascites and cachexia, even many years after the rupture has occurred. A sudden onset of bile colored ascites in an otherwise healthy person, regardless of the fact that no hepatic cyst is visible, should raise the suspicion of intraperitoneal rupture in endemic areas.

The diagnosis is established with the use of US and CT scan as imaging methods. The presence of intra-abdominal fluid and the detached membrane of the hydatid cysts are clearly demonstrated. Diagnostic peritoneal lavage has been also considered to be a helpful and highly specific test for hydatid cyst perforation.

### Management

Emergent operation is mandatory to remove the intraperitoneal fluid and eradicate the cysts. The most important steps during the surgical treatment are irrigation of the peritoneal cavity with a sufficient amount of scolicidal agents, removal of all cystic contents from the peritoneal cavity, and combined with meticulous peritoneal washing. An important parameter influencing the choice and duration of any surgical intervention is the general hemodynamic status of the patient. Furthermore, the post-operative administration of benzimidazole carbamates (mebendazole and albendazole) for a period of 3–6 months is arbitrarily advised.

### The Rupture in the Thorax<sup>[18-21]</sup>

Thoracic complications of hepatic hydatid cysts result from the proximity of hydatid cysts in the liver and the diaphragm and are seen in approximately 0.6–16% of cases. Several factors, such as pressure gradient between thoracic and abdominal cavities, mechanical compression and ischemia of the diaphragm, sepsis in the hepatic cyst, or chemical erosion by bile, participate in promoting intrathoracic evolution of hydatid cysts of the hepatic dome. Pathognomonic clinical findings are bile-stained sputum, hydatid vomica and hydatidoptysis sputum containing daughter cysts, sometimes bile stained. In selected cases, respiratory distress syndrome may also be encountered. When the rupture involves the pleural cavity, pneumothorax and systemic anaphylactic reaction are usually present.

The imaging work-up includes a chest X-ray (CXR), US, and in fewer cases CT or MRI scans. When a diaphragm with blurred or erased limits is depicted in the CXR, US are then performed, which shows the hepatic hydatid cyst and its close proximity to the thoracic lesions. The diagnosis is established with the demonstration of the diaphragmatic discontinuity. CT and MRI scans can offer additional information with regard to the precise connections between the hepatic hydatid cyst and the intrathoracic lesions.

### Management

Depending on the degree of cyst evolution and involvement, there are five surgical grades of diaphragmatic or transdiaphragmatic thoracic involvement in hepatic hydatid disease. Grade 1 represents firm adherence between the diaphragm and the cyst surface without diaphragmatic perforation. In Grade 2, the cyst perforates the diaphragm, but there is little invasion of the thoracic cavity. Cyst perforation through the diaphragm with either cyst growth inside the thoracic cavity or daughter vesicle formation is Grade 3. Disease of the lung parenchyma by either cyst connection with the bronchial tree or compression and atelectasis of the lung is considered Grade 4. Establishment of a bronchobiliary fistula is Grade 5.

Surgery remains the main therapeutic option. During surgical operations, the treatment of the liver cyst, hepatodiaphragmatic disconnection, treatment of intrathoracic lesions, restoration of the diaphragm, and a secure biliary tract represents the most important goals. Thoracotomy must be performed in cases of intrathoracic collection. Laparotomy is necessary when the biliary tree is involved and, therefore, requires adequate drainage. An isolated abdominal approach is considered sufficient when there is direct rupture into the bronchial tree.

### Liver Hydatid Cysts Ruptured into the Pericardium<sup>[2,22,23]</sup>

The parasites that escape through the hepatic veins may reach the right atrium, and, consequently, enter the coronary arteries' circulation. As a result, they may be situated in any part of the myocardium. A rupture into the pericardium results in disseminated pericardial echinococci, acute cardiac tamponade, and chronic constrictive pericarditis. Nearly 10% of all cases of myocardial hydatid cystic disease rupture into the pericardium, and most of these events are fatal.

Initially, patients can be asymptomatic (26.5%). They can also experience certain symptoms due to myocardial compression, or even, dyspnea, cough, fever, and hemoptysis.

At present, the most valuable imaging modalities for detecting cardiac echinococcosis are echocardiography and CT scan, which give appropriate information regarding its size and relation to nearby structures.

### Management

Cystopericystectomy is the "gold standard" surgical procedure, but it is sometimes unsuitable for particular sites, which should be treated with partial pericystectomy. The operative mortality rate ranges from 0% to 5%.

### Rupture into Other Cavities or Organs<sup>[24,25]</sup>

Although rupture to the gastrointestinal tract is rare, it has been described, specifically to the stomach and the duodenum. Rupture to these organs presents with abdominal pain, nausea, and vomiting. Radiologic studies with CT tomography reveal the existence of air inside the echinococcal cyst, and communication of the cyst with the stomach or the duodenum. The diagnosis is confirmed with endoscopy. Treatment of these cases is surgical consisting partial or total pericystectomy and suture of the viscus.

### Rupture to Major Vessels (Portal Vein, Inferior Vena Cava, and Aorta)<sup>[26,27]</sup>

The rupture of echinococcal cysts of the liver to the hepatic veins or the inferior vena cava can lead to the spread of hydatid disease to the lungs or to the pulmonary arteries. Symptoms include cough, hemoptysis, and dyspnea. In endemic areas, there must be increased clinical suspicion in patients presenting with these symptoms, especially if they have undergone liver surgery for hydatidosis. Inferior vena cava thrombosis has been reported as well.

Most infrequent complication of hydatid cysts of the liver is rupture to the portal vein. Four cases have been reported in the literature. In this situation, symptoms include abdominal pain, fever, and signs of portal hypertension. Anaphylactic shock is expected after rupture of a cyst in vessels.

### Cutaneous Fistulization of Liver Hydatid Disease<sup>[2,28]</sup>

Cutaneous fistulization is a rare but serious complication of hydatid disease involving many organs, including the liver. Hydatid cysts follow several stages before reaching the abdominal wall to develop into an external rupture. Stage I hydatid lesions protrude into the innermost muscular layer of the abdominal wall. Stage II lesions pass beyond the muscular layer and protrude into subcutaneous soft tissue. Stage III is characterized by the passage of lesions into subcutaneous tissue and their fistulization in the skin, which is called an external rupture, external fistulization, or cutaneous fistula. A typical clinical sign of cutaneous fistulization in a patient presenting from an endemic area is discharge of hydatid fluid or daughter vesicles from the external orifice of a fistula. Combination of clinical presentation, serologic tests and radiological workup can lead to final diagnosis easily in almost all patients. Useful radiologic tools include US, CT, and MRI, whereas ERCP or PTC is invasive methods used for both diagnostic and therapeutic purposes. The most useful radiological method for cutaneous manifestations of hydatid disease is contrast-enhanced fistulography. This technique is helpful in specifying the extension of the fistula, the location and size of the fistulized lesion, and its relationship with bile ducts. Success rate of fistulography in demonstrating organ involvement of a cysto-cutaneous fistula is 85.7%.

### Management

In terms of appropriate treatment pathway, the best approach is a combination of medical treatment and surgical intervention. A 2–4 week neoadjuvant medical treatment followed by elective surgery is considered to be the safest pathway in non-urgent cases. Surgical treatment includes *en bloc* resection of the primary hydatid lesion causing the complication, diseased skin region, and fistula tract.

### Management of Echinococcal Cyst Communication with the Biliary Tract (Rupture to the Biliary Tract)<sup>[2,10,29-32]</sup>

The management of communications of the echinococcal cyst with the biliary tract represents one of the most critical steps in the conservative surgical management of hepatic hydatidosis. The rupture in the biliary tract represents an independent

parameter that increases post-operative mortality and morbidity in a statistically significant degree, while insufficient management leads to life-threatening complications. Sometimes, the communication with the biliary tract can be very easily identified. It is possible that a single hydatid cyst can have multiple fistulas with the biliary tree, while, similarly, a patient with multiple cysts, biliary communication can be present in more than one cyst. Reconstruction and management are both feasible and successful in most cases. However, in cases with communication of the cyst with major biliary branches, management can be quite challenging.

Pre-operative localization of most fistulas with the biliary tract is not always possible. Symptoms (cholangitis, obstructive jaundice, and biliary colic), as well as pathologic laboratory exams (liver dysfunction, increased cholestatic enzymes, and dilated biliary tract) are only present in a small percentage of patients (16% and 18%, respectively). Predisposing factors that may warn the surgeon for existence of communication with the biliary tract are: Advanced age of patient (with a long history of echinococcal disease of the liver), significant size of cysts, multiple cysts, as well as bilateral lobe cyst localization. The management of fistulas depends on a lot of factors: (1) The number of cysts, (2) cyst type, (3) localization of the cyst, (4) size of the fistula, (5) biliary branch involved, (6) patient clinical status, (7) liver function and, finally, and (8) surgeon's experience.

Bilus content of the hydatid cyst is highly indicative of fistula with the biliary tree. In most cases, the exact orifice of communication is not visible intraoperatively. In these cases, a detailed inspection of the internal surface of the cysts should be performed after wide excision of the pericyst. A suitable maneuver is the placement of a gauge in the cyst, while the surgeon applies pressure on the gallbladder. The gauge is then inspected for bilus spots.

Since sometimes the precise localization of the bile fistula is impossible intraoperatively, certain authors suggest the use of intra-operative cholangiography, in which daughter cysts can be revealed in the biliary tree. However, routine use of intra-operative cholangiography is avoided for the following reasons: (1) Only a small percentage of patients present concretions of hydatid contents inside the biliary, and in these cases an intra-operative ultrasound would be equally helpful, (2) in patients reporting cholangitis symptoms, endoscopic retrograde cholangiography has already been performed, and (3) intra-operative cholangiography can reveal contrast outside the biliary tree, confirming the existence of biliary fistula, without being able to locate the exact point of leakage. An alternate method used, is the infusion of methylene blue inside the gallbladder or bile duct. This method can reveal very small communications that would have been missed by the surgeon. The disadvantage of the method is diffuse spread of the dye in the entire cyst cavity.

The reconstruction of the biliary fistula should be as simple as possible. Reconstruction methods can be divided in the following categories: (1) Suturing of the communication (simple suturing, suturing with simultaneous placement of T-tube common bile duct drainage, intralateral pericystectomy, and caputotomy), (2) internal drainage procedures (biliodigestive bypass, transduodenal sphincterotomy, and internal transistular drainage with or without transduodenal sphincteroplasty), (3) external drainage procedures (bilateral drainage, and cystobiliary disconnection), (4) reconstruction procedures

(pericystojejunostomy, intracavitary biliodigestive bypass, or bile duct repair), and (5) hepatic resections.

In the majority of patients, the communication of the hydatid cyst involves a small peripheral branch of the biliary tract, therefore simple suturing of the communication is sufficient. In this case individual absorbable sutures should be used.

T-tube can be used as safety for decompression in cases of suturing of a large fistula between a stiff pericyst and big biliary branch, or in cases where the bile duct has been opened for the extraction of hydatid contents.

In rare cases, the orifice of the fistula is not suitable for simple suturing. In these cases, combined drainage techniques should be performed, for the fistula to resolve automatically. Drains should be placed inside the pericyst cavity, and the biliary tract should be decompressed with internal or exterior drainage. The bilateral drainage includes the placement of drains in the pericyst cavity, while the bile duct is drained with a T-tube (Kehr drainage). Increased post-operative bile leaks have been reported by this method leading to increased hospital stay. The Permodo procedure consists of the interruption of the communication between the hydatid cyst and the biliary tract with an anatomic separation between the residual cavity and the biliary fistula. A catheter with multiple orifices at both sides is placed through the orifice of the fistula for drainage of the fistula, while one similar is placed inside the cavity of the cyst for drainage of the cyst. The bile duct is drained with a T tube.

In the internal drainage procedures, the decompression of the biliary tract is achieved with: (1) Endoscopic sphincterotomy, (2) transduodenal sphincteroplasty, and (3) biliojejunal anastomosis. Internal drainage through the fistula is accomplished after the meticulous evacuation of hydatid contents and suturing of the pericyst. The pericyst will be drained in the biliary tract through the existing fistula.

The indications for performing a cystojejunostomy are very rare and concerns large and centrally located cysts with communication with a major biliary branch with an orifice that is not suitable for suturing.

Biliojejunal bypass is performed in cases with massive entry of hydatid contents and daughter cysts in the biliary tract, in acute cholangitis, as well as in cases where the bile duct is dilated.

In cases of extensive communication with the biliary tract, the ideal approach includes radical surgical treatment (total or partial pericystectomy, and hepatectomy). In these cases, radical surgical treatment ensures less post-operative complications and smaller hospitalization.

## CONCLUSION

Complications deriving from liver hydatid disease can easily be distinguished from silent hepatic hydatidosis due to distinct clinical and radiological features. However, there is necessity for meticulous preoperative planning, as sometimes the surgical management includes complex maneuvers. Moreover, complicated liver hydatid cysts are characterized by increased frequency of post-operative complications which subsequently lead to prolonged post-operative period.

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