



(RESEARCH ARTICLE)



## Clinical presentation, diagnosis and surgical treatment of intrabiliary ruptured hydatid disease of the liver

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### Abstract

**Background:** Intrabiliary rupture (IBR) of liver hydatid cyst is common and serious and fatal complication. The incidence of (IBR) varies from 1% to 25%. The treatment of IBR is still controversial among surgeons. We aimed to show clinical presentations, diagnosis treatment and outcome of IBR in liver hydatid cysts by reviewing our cases.

**Method:** In a retrospective study we collected data of records of all patients who were operated on for hydatid disease of the liver operated between 2016 and 2021 at Razi teaching and private Arya Hospital were reviewed. 322 patients who had been operated for hydatid cyst of the liver were detected. Approval for this study was obtained from our Institutional Review Board.

**Results:** Intrabiliary ruptured hydatid cysts of the liver was determined in 14 of the patients was in frank IBR and 32 patient with occult IBR was determined in 322 patients was operated for hydatid cysts of the liver. The average age of patients was 28 to 64 years (range 21 - 68 years). The most frequent symptoms were right upper quadrant, epigastric pain, dyspepsia, jaundice, fever, weight loss and pruritus

**Conclusions:** Intrabiliary ruptured hydatid cysts of the liver is the most common complication. With appropriate evaluation and intervention, this will lead to decreased postoperative bile leakage and fistula formation, a major cause of morbidity and mortality. Every effort should be made to detect and prevent these complications, ERCP and MRCP can help for diagnosis and treatment of IBR. Laparotomy, choledochal exploration should be performed during operation. For decompress intrabiliary pressure T-tube drainage may be preferred in the management of intrabiliary ruptured hydatid cysts because of decreased morbidity, the ability to, easier monitoring of the biliary drainage.

**Keywords:** Liver; Surgical treatment; Hydatid disease; Diagnosis; Intrabiliary rupture

### 1. Introduction

Hydatid cysts, also known as echinococcosis, is a serious and problematic health issue worldwide, and this parasitic disease is widespread in most West Asia, Mediterranean countries, New Zealand, South America, the Far East, Australia,

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and East Africa [1–3]. Humans are intermediate and incidental hosts and are infected either the direct route via exposure to contact with infected dogs or other canines or indirect routes via consumption of food, water, and infected material on the ground especially vegetable [2–4]. Biliary cirrhosis may also be a danger sequel of intrabiliary rupture of liver hydatid cysts because of clostridia [5–7]. The majority of patients show an single organ involvement as brain, spleen or kidney with a single cyst, and 75–85% of cysts are localized in the liver of patients and 15 to 25% is lung and concomitant of liver and lung in 5 to 10%, all or gan of body can infected [2, 5]. Some cysts may grow at an average rate of 1–20 mm per year, and these patients usually asymptomatic and survive with no evident changes for a long time; other cysts can become calcified and completely disappear and some undergrow sequestration [5, 8]. An enlarging cyst may compress and cause atrophy and fibrosis of the liver and produce biliary cirrhosis [5, 9]. The compression and displacement of biliary ducts can frequently produce a spontaneous rupture in biliary ducts [7, 21]. Intrabiliary rupture (IBR) is the most common and serious complication of hepatic hydatid cyst (HHC) [5, 10, 12]. The incidence varies from 1% to 25% although an incidence of 64.75% has been reported from a multicentric study in Tunisia [7–21]. Early diagnosis and therapy are mandatory in the case of an intrabiliary rupture of a liver hydatid cyst, which can result in the obstruction of the biliary duct with 50% mortality [4–7, 14]. Imaging tools, such as ultrasonography (U/S), abdominal computed tomography (CT), magnetic resonance cholangiopancreatography (MRCP), and endoscopic retrograde cholangiopancreatography (ERCP), are useful devices to diagnose the disease [2, 8, 9, 17]. U/S and CT scans are the first diagnostic tools of choice and can be applied under all conditions [1, 2, 17, 18]. Of the more invasive instruments, ERCP can help establish definitive detection and treatment with sphincterotomy in patients affected by intra-biliary rupture of a cyst, and MRCP can diagnose the site of the obstructions of the biliary system [2, 8, 9, 15, 17, 18]. This study report show the clinical presentation patients with liver echinococcosis, the various complications of a hepatic hydatid cyst, especially intrabiliary rupture, and the methods of diagnosing and managing IBR. Although there is some problem on the medical and surgical treatment of uncomplicated hydatid cyst but the treatment of intrabiliary rupture still remains controversial.

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## 2. Material and methods

The record 322 cases of liver hydatid cyst were analyzed *retrospectively*. Case records of patients who were operated on for hydatid disease of the liver diagnosed between January 2016 and December 2001 at Razi and Arya hospital were searched and 322 patients who had been operated for hydatid disease of the liver were detected. Of these, 14 patients (4 females, 10 males) with frank IBR and 32 patients with occult (Male 20, female 12) were retrospectively reviewed for intrabiliary ruptured hydatid disease of the liver. Occult IBR was diagnosed in the operation time with aspiration of bile-stained cystic fluid (Figxx). Patients were evaluated according to age, sex, clinical findings, cyst number and stage, abdominal ultrasonography, CT-scan and MRCP, ERCP, frank or occult IBR, surgical methods, complications, morbidity and mortality and outcome.

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## 3. Results

Intrabiliary ruptured hydatid disease of the liver was determined in 14 of the patients was in frank IBR and 32 patient with occult IBR was determined in 322 patients was operated for hydatid disease of the liver. The average age of patients was 28 to 64 years (range 21 - 68 years). The duration of the symptoms was 14 to 21 month (range 3 to 18 month). The most frequent symptoms were right upper quadrant, Epigastric pain, dyspepsia, jaundice, fever, weight loss and pruritus. Diagnosis of hydatid cyst was principally made using ultrasonography, CT-scan, MRCP and ERCP (Fig1, 2, 3). In frank IBR, eight cysts were located in the right lobe, four in the left lobe, and two in both of right and left lobes. In occult IBR, 18 cysts were located in the right lobe, 8 in the left lobe, and 6 in both of right and left lobes. The size of the cysts in frank and occult IBR was 14 to 8 cm (range 8 to 18 cm). The average diameter of the common bile duct (CBD) in frank IBR was 30 to 2/5 mm (range 20 to 50 mm). In occult IBR The average diameter of the common bile duct was in normal range, and according to Gharbi and the World Health Organization classification 18 in class one, 8 in class two and 6 in class three [16]. In frank IBR dilated of CBD and daughter cysts, laminated membrane and debris in the CBD in in all 14 patients were found during operation but in the occult IBR needle aspiration during operation show bilious fluids. Before operation we usually used 100mg hydrocortisone IV injection in all patients underwent operation and choledochotomy, evacuation and lavage of the CBD was performed and evacuation remnant cavity irrigated with saline 5% (4). CBD irrigated with saline normal and evacuated all debris, daughter cysts and laminated membrane next Partial cystectomy and capitonnage were performed in 8 patients, omentoplasty 6 patients plus T-tube drainage in all patients (Fig4, 5). In addition (2, 3) In occult IBR Partial cystectomy and capitonnage were performed in 22 patients, omentoplasty 10 patients. In addition in 12 patients internal opening of the biliary fistula was found in the cavity post evacuation and sutured in. In all case a Foley catheter put in the cavity after omentoplasty, before closure laparotomy a pancreas drain put in the subhepatic region and partial cystectomy (Fig 5). Wound infections developed in 6 patients, pulmonary complication in 8 patients as pneumonia, atelectasia, effusion, empyema. Biliary leakage in 5 patients, leakage stopped in three patients and

two patients with a high output more than 400ml biliary fistula occurred on the first post-operative day and continued for a week. ERCP and sphincteroplasty were performed and the biliary fistula stopped at the 4th post-operative week. One patient died from sepsis, multiple organ failure and hepatic failure. The average period of hospitalisation was 16 to 8 days (range 8 to 12 days). T-tube drainage was removed after T-tube cholangiography (Fig 6).

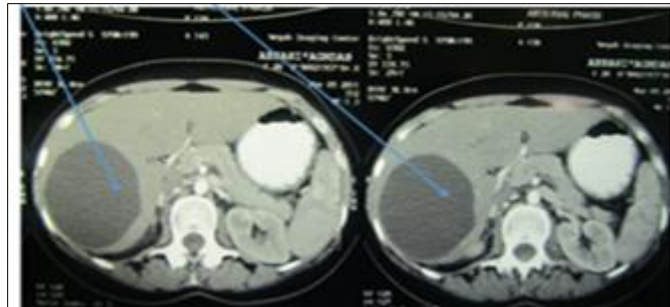
#### 4. Discussion

Intrabiliary rupture is a common and serious complication of hepatic hydatidosis. It occurs especially in centrally localized and high stage cyst. High intracystic pressure up to 80 cm H<sub>2</sub>O is a predisposing factor. The cyst rupture can occur in three clinical forms (3,23). Contained rupture occurs when the cyst contents are confined within the pericyst. Communicating rupture defines tearing of the pericyst and evacuation of cyst contents into the biliary tract or bronchioles. Direct rupture describes complete tear of the cyst wall and spillage of the cyst contents into the peritoneal or pleural cavity. Small cysto-biliary communications develop in 80-90% of all HHCs [1,2,3,4]. Furthermore, there are two different clinical settings associated with intrabiliary rupture: frank intrabiliary rupture and simple communication. In the former, the cyst content drains to biliary tract and causes cholestatic jaundice. In the latter simple communications are frequently overlooked and could cause biliary fistulae post-operatively [3,4]. If the cystobiliary opening was less than 5 mm, spontaneous drainage of the cystic content was uncommon and could be treated by suturing under the direct vision [15,19,20]. If the CBD diameter was larger than 5 mm, cystic content migration into the biliary tract would occur in 65% of the cases [9,16]. Vesicles, debris and purulent materials may be found in the biliary collection [21]. In all patients the most frequent symptoms were right upper quadrant pain and flatulence [3,4,21]. Obstructive jaundice and fever have been recorded in 90% and 20% of the cases respectively. Nausea and vomiting were rare [8,9,12,21]. Diagnosis of IBR is difficult and can be established pre-, intra- and post-operatively. When obstructive jaundice is present, US, CT-scan, magnetic resonance imaging (MRI) and scintigraphic investigation can show the cyst and cystobiliary communications, but in patients with no jaundice, a correct diagnosis can be made in only 25%. Radiodiagnostic evaluation can also demonstrate cystic content in the gall bladder and the CBD [9,12,14,16]. On the other hand, cholelithiasis and choledocholithiasis are common (81-61.53%) coincidental diseases [6,9,12].

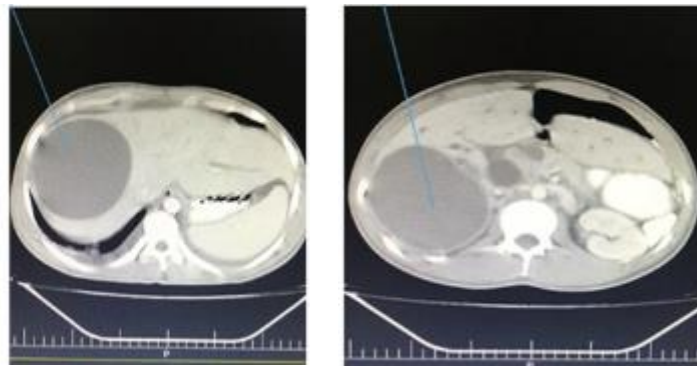
Serological tests can also be helpful for diagnosis (7,9). We don't use this test in our patients. If obstructive jaundice was not present and cystobiliary connection could not be seen pre-operatively in laboratory, three findings should raise suspicion of cystobiliary fistulas, bile-stained cystic fluid during operation, exposure of bile leak orifice intra-operatively; observation of CBD dilatation intraoperative, presence of cyst elements in the CBD; postoperative bile drainage from the cavity drains [24,25,26,27]. Bile leakage may be seen in 11.7-17.07% of the cases during the operation [24,25,28]. In these cases, cholangiography could be done by cystic ductus or from puncture of CBD and injection of radioopaque solution or methylene blue diagnose intrabiliary rupture or to see the orifice of leakage ([12,17,18,28

Eleftheriadis emphasized that intraoperative choledochoscopy can be helpful in the diagnosis of IBR [29]. When an IBR is diagnosed preoperative or during operation, there are need treatment with surgery of hepatic hydatid cyst (3,4,5,7). Cystic material should be evacuation, removal of laminated membrane and daughter cysts, irrigation of cystic cavity are necessary (11,12,13,21). If orifice leakage site is seen and no cystic content is observed in a normal size of CBD, suturing the orifice is sufficient to prevent fistula (26, 27, 28). Videolaparoscopic suturing of the orifice has been reported in literature [30]. In our case we don't use this procedure. When cystic content was seen in a normal CBD, choledochotomy, evacuation and debris from biliary system irrigation of CBD with 0.9% NaCl solution and T-tube drainage are enough (2,4,21). If CBD diameter is enlarge with cystic content in it or in the gallbladder, choledochoduodenostomy is preferable (7,21). We never do this procedure in our patients but cholecystectomy perform. Some authors have reported the the large size of T-tube drainage in cases with high morbidity rates are need [3,4,21,30,31,32]. Roux and Y hepaticojejunostomy have been reported for the treatment of bile duct with stricture associated IBR [28,29,34]. In our study we didn't see such case. Open sphincteroplasty was also used in the last decades [35]. We didn't do such operation. The presence of cholangitis has been reported in patients even with T-tube drainage because it could cause cholestatic icterus [2,4,21,27] and is also a source of infection [28]. Some authors have used cystojejunostomy because cystic content may drainage into the biliary tract could result in obstructive jaundice and cholangitis [21,34]. When IBR was occurred during the surgical treatment, biliary fistulae (up to 1 000 mL/d) if this occurred ERCP would be necessary (17,18). Post-operative biliary fistula rate was about 20% in all cases [22,23,30,31]. In the cases of cystobiliary fistulae, transsphincteric evacuation of the CBD and sphincteroplasty could be applied. Nasobiliary drainage could also be done. Usually, the majority of biliary fistulae could be closed in a few weeks [7,8,9,21,34]. The usage of endoprotheses in biliary fistula was not common (21,36). This can be considered in high output bile leakage or for fistulae was not close with every treatment [31]. The morbidity and mortality rates of all patients were 19.44-43.03% and 1.8-4.5% respectively in literature (21,28,31). In our study one of patient died. The most common causes of deaths were sepsis and hepatic failure [21,34,35]. Hospital stay was the longest in the T-tube group. The patients undergoing choledochoduodenostomy had longer hospital stay than those undergoing simple

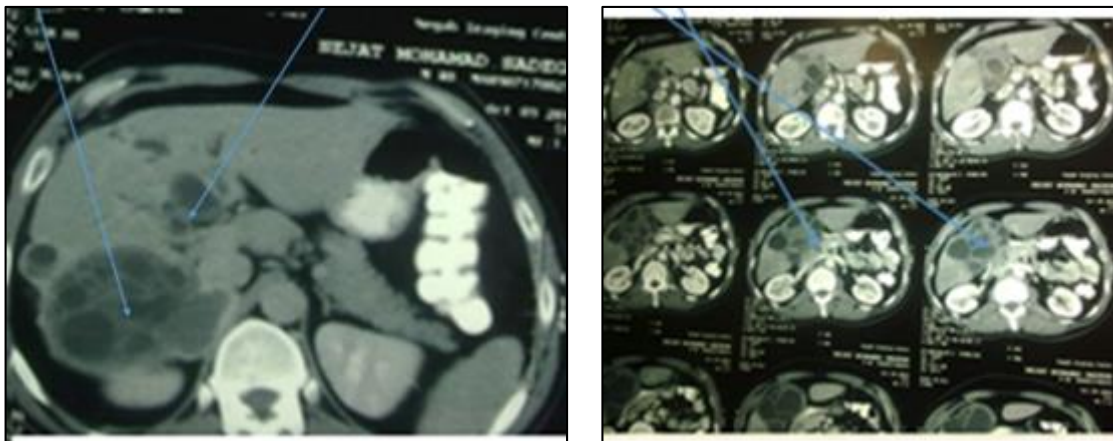
orifice suturing[21,28,29,34]. In conclusion, IBR has an algorithm in the diagnosis and treatment (34). If it is not detected pre- or intra-operatively, a biliary fistula is common, Wound infections developed in 6 patients, pulmonary complication in 8 patients as pneumonia, atelectasia, effusion, empyema. Biliary leakage in 5 patients, leakage stopped in three patients and two patients with a high output more than 400ml biliary fistula occurred on the first post-operative day and continued for a week. ERCP and sphincteroplasty were performed and the biliary fistula stopped at the 4th post-operative week. One patient died from sepsis, multiple organ failure and hepatic failure. The average period of hospitalisation was 16 to 8 days (range 8 to 12 days). T-tube drainage was removed after T-tube cholangiography (Fig x).



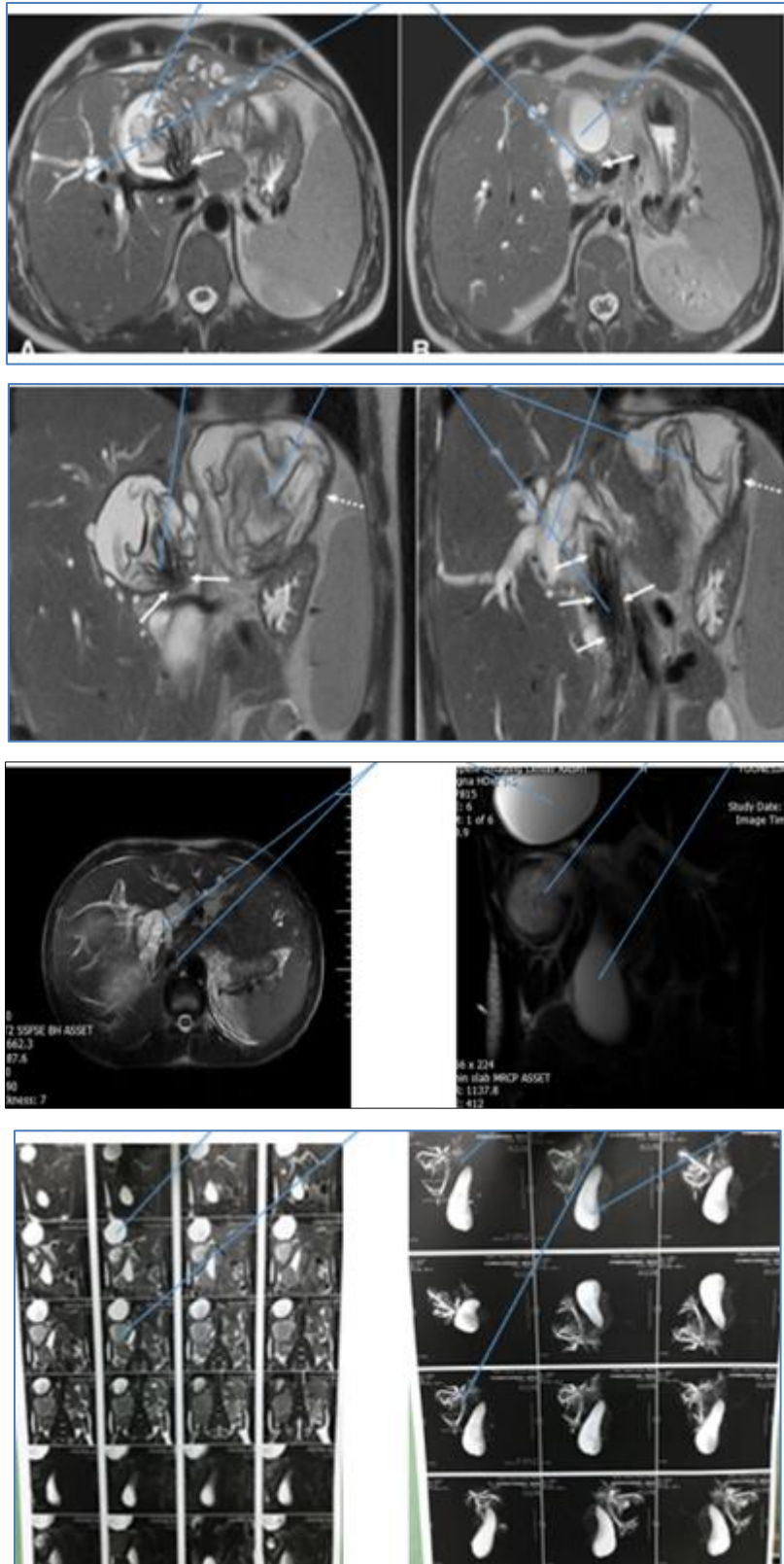
**Figure 1** Intact liver hydatid cyst in CT-scan



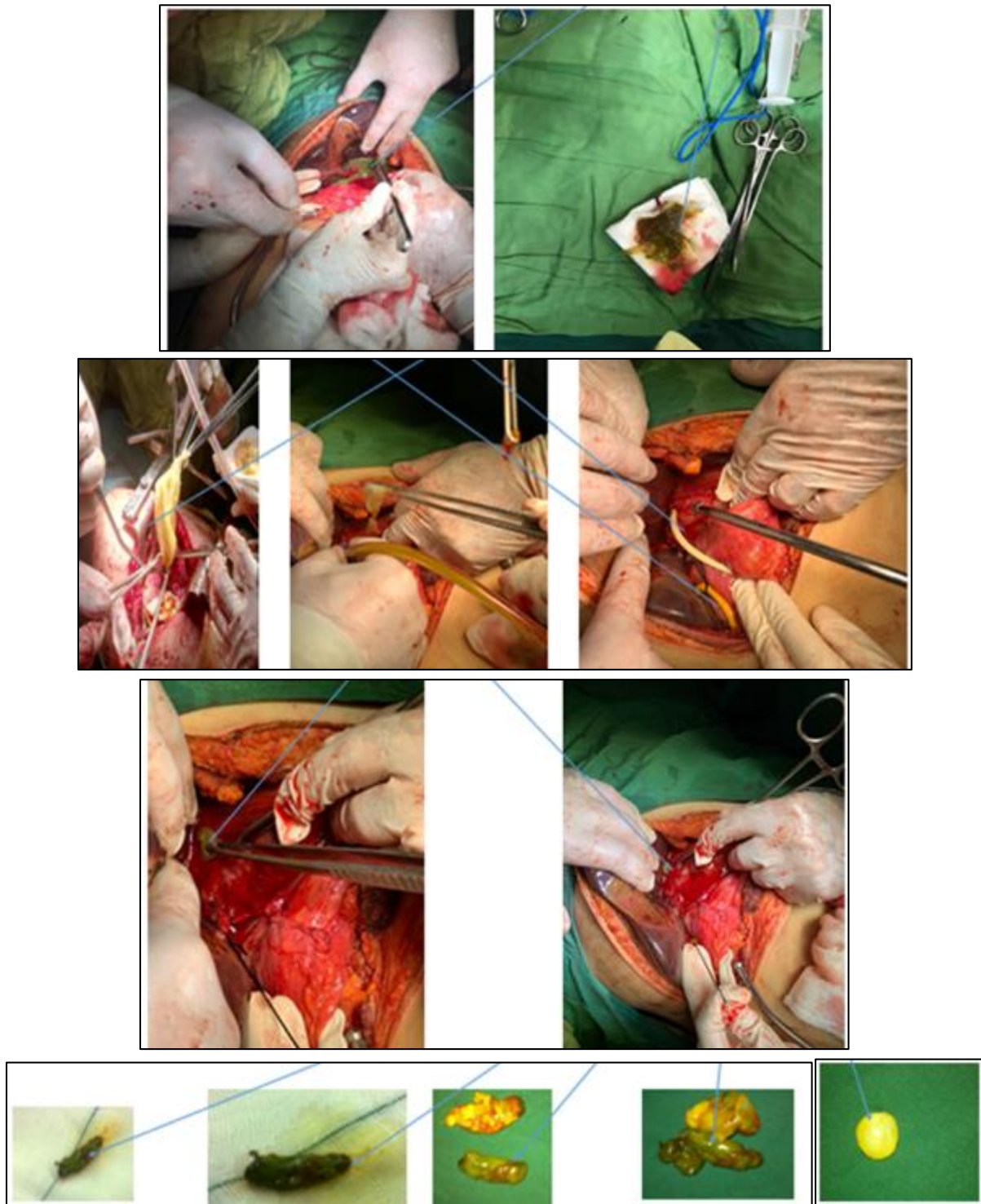
**Figure 2** Small rupture liver hydatid cyst in CT-scan



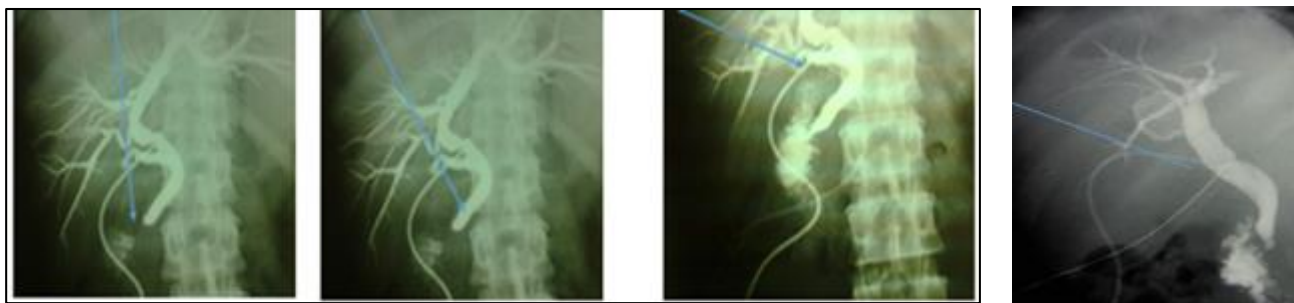
**Figure 3** Large rupture of liver hydatid cyst in CT-scan



**Figure 4** Intra biliary rupture of liver hydatid cyst in MRCAP Fig5 show cyst element during operation



**Figure 5** Paratomy shwo T-tube,external drainsge,doughtercyst and laminated membrane



**Figure 6** T-tube cholangiography

## 5. Conclusion

Intrabiliary ruptured hydatid cysts of the liver is the most common complication. With appropriate evaluation and intervention, this will lead to decreased postoperative bile leakage and fistula formation, a major cause of morbidity and mortality. Every effort should be made to detect and prevent these complications, ERCP and MRCP can help for diagnosis and treatment or IBR. Laparotomy, choledochal exploration should be performed during operation. For decompress intrabiliary pressure T-tube drainage may be preferred in the management of intrabiliary ruptured hydatid cysts because of decreased morbidity, the ability to, easier monitoring of the biliary drainage.

## Compliance with ethical standards

### *Acknowledgments*

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### *Disclosure of conflict of interest*

No conflict of interest to be disclosed.

### *Statement of ethical approval*

The study was performed in accordance with the declaration of Helsinki and approved by the Ethics Committee of Guilan IR.GUMS.REC.1397.151.

### *Statement of informed consent*

There are no conflicts of interest.

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