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Presentations, diagnosis, treatment and outcome of complicated liver hydatid cysts: from an endemic area

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Abstract

Background: Hydatid cysts (HC) can involve all organs of the human body but most often in the liver (77% to 50%) and lungs (35% to 18%). (HC) is endemic in certain areas of the world as in the Middle and Far East, Iran, Australia, New Zealand, and South America. (HC) remains a considerable public health problem.

Methods: In a retrospective study was carried out in north of Iran Guilan Province. A total of 322 patients who were operated on for 382 (HC in the liver between 2016 to 2020) were evaluated. Patients were divided into two groups: complicated (HC) in 87 patients and uncomplicated (HC) in 235 patients. Frank intra-abdominal in 15 and frank intrabiliary rupture in 14, occult intrabiliary rupture in 32, cyst infection in 15, intrapleural and intraparietal rupture in 10 patients.

Results: The patient's gender included 72% were men and 28% were women with a median age of 18 to 64 years. Abdominal ultrasound, computed tomography, MRCP and ERCP were the diagnosis tools. In interpleural or intraparietal rupture thoracotomy and laparotomy performed with evacuation and omentoplasty, capitonnage and decortication. In intracardiac rupture thoracotomy and pericardiectomy and laparotomy and omentoplasty, pericystectomy or capitonnage was performed. There was one mortality. The postoperative morbidity was in 28 patients in complicated cysts and 12 in uncomplicated cysts.

Conclusion: Complicated hydatid cyst of the liver can be successfully managed surgically with good long-term results. Complicated liver echinococcosis demonstrates several distinct features that differentiate it from the noncomplicated disease. Frequently severe clinical manifestations, complexity of surgical management, and the increased postoperative complications characterize complicated liver echinococcal disease.

Keywords: Liver hydatid disease; Complicated liver hydatid; Drainage; Operative techniques

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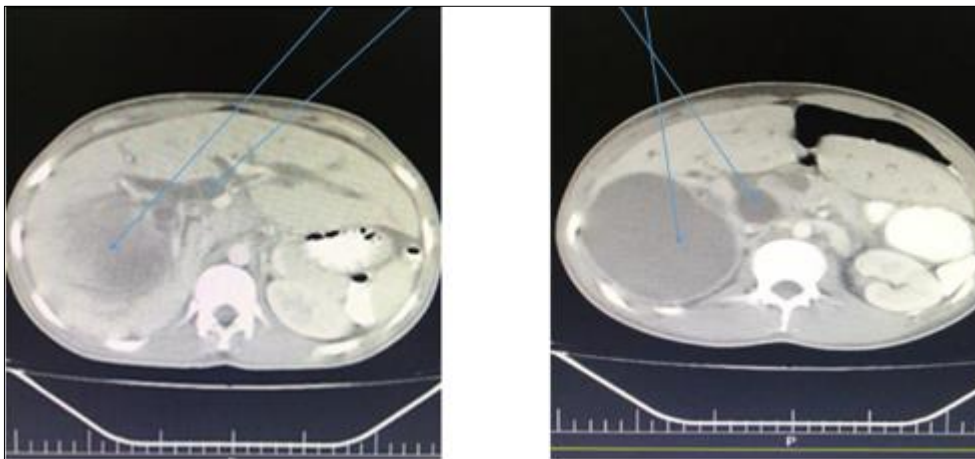
1. Introduction

Hydatid disease is a serious health and infection problem in world especially in endemic countries. There are reports from several countries, the disease is endemic in the Mediterranean region, Far East, South America, and Middle East such as Iran [1, 2, 3, 4]. It is a parasitic infection caused by the *Echinococcus granulosus* [1, 2]. Humans are accidental infected and do not have any role in the life cycle of hydatid cysts [2, 3, 4]. Humans are infected by eggs of this parasite from vegetable, soil or water which contaminated by the feces of dogs [1, 2, 3, 4]. Hydatid cysts may involve all organ in the body but occur most frequently in the liver (50%–77%) and lungs (18%–35%), and occasionally in other organs [1, 2, 3, 4, 5, 6]. Concomitant liver and pulmonary hydatid cysts occur in 4%–25% of patients [1, 4]. In some time, Hydatid cyst is a chronic disease with late presentation and often is asymptomatic (2, 3, 5). It may be found during routine clinical examination and serologic, radiographic, or ultrasonographic screening [5, 6, 7, 9]. The clinical signs and symptoms of hydatid cysts depend on locations in the involvement of organs (deep or superficial, right or left lobe), size, adjacent organs, and complications such as infection or cyst rupture in the peritoneal cavity, intrabiliary, in the pleural or lung parenchyma and rarely intracardiac cavity [1, 2, 4, 5, 8]. Secondary bacterial infection of the hepatic hydatid cyst is rare complication, Essential prerequisite for the bacterial infection of the cyst is the rupture of both pericyst and endocyst, so bacteria can enter the cyst [10]. The presence of air forming an air fluid level inside the cyst, while the size of the cyst remains not change, this finding often observed on CT in such cases and may indicate the infection of the cyst, this pattern may be seen in other cases in the liver [11]. The clinical status of the patient imagine finding will contribute to the final diagnosis in cases of infection of the cyst which present as a hepatic abscess [10–11].

A cyst may rupture into the biliary systems, into the hollow organ, through the diaphragm into the pleural cavity or pulmonary parenchyma, or directly in the peritoneal cavity [4, 5, 6, 8, 12]. Rupture of the cyst may occur after trauma or spontaneously due to suddenly increased intracystic pressure or during treatment with Albendazole [4, 6, 9]. Complications of hepatic hydatid cysts are rupture in the peritoneal cavity or interbiliary tract and secondary bacterial infection [4–6, 7, 8, 9]. A hydatid cyst rupturing into the peritoneal cavity or may cause some symptoms including abdominal pain, urticaria, anaphylaxis, and sudden death or shoulder pain due to irritation of diaphragm [7–8–9]. Presentation is usually dramatic with acute abdominal signs, such as guarding, rebound, and tenderness [1, 3, 5, 7, 8, 9]. Therefore, rupture of hydatid cyst requires both emergency surgery and careful postoperative care and anti-allergic medication as hydrocortisone and epinephrine in emergency room before surgery. In this case series, we want to show clinical presentation, diagnosis, surgical approach, mortality, morbidity and recurrences of patient who had hydatid cyst of liver with complications.

2. Materials and Methods

In this study all Patients with liver hydatid cysts who were treated by surgery between January 2016 and January 2020 were retrospectively evaluated. Records of clinical presentation and examination, diagnosis tools, history, abdominal ultrasound (US), computerized tomography (CT), (MRCP) and ERCP (Figs 1). Liver and renal function and CBC tests was evaluated. Blood eosinophils have very low sensitivity. In our Hospitals we don't use serologic testes for diagnosis.



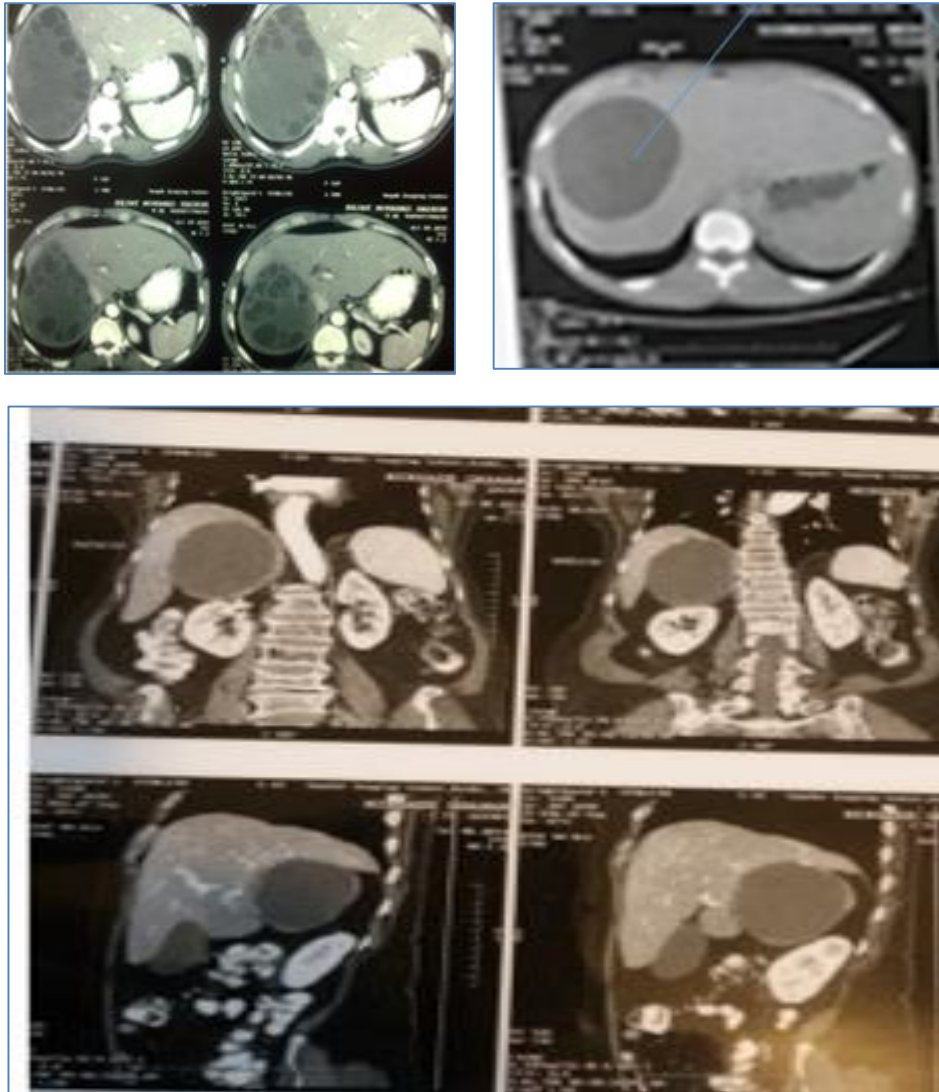


Figure 1 CT and MRI of complicated and uncomplicated liver hydatid cysts

The patients were divided into two groups, complicated hydatid cyst as frank or occult rupture(14-32) and uncomplicated hydatid cyst (UCHC) in 250 case. Patients if presented preoperative with the following symptoms: jaundice and possibility of cholangitis diagnosis of intrabiliary rupture, preoperative cough with sputum, dyspnea and chest pain diagnosis of intrabronchial Or intra paranchimal rupture, Abdominal pain, tenderness and guarding diagnosis of intera peritoneal cavity perforation.these type of compilations named frank cyst rupture intraoperative cyst aspiration show bile-stained or purulent content this type named occult rupture.If patient present with RUQ pain and fever these type is infected cysts. The remaining cases were in group of UCHC.

All liver hydatid cysts were indicated for surgery except uncomplicated multiple small size cysts, cysts smaller than 5 cm in diameter, Cysts located deep in the liver parenchymal or liver cysts with multiple intraperitoneal cysts, these kind of cyst managed with Albendasol and six month interval imaging studies. Pricyest layer with thick calcified cysts were managed with observation because these kind of cysyts usually are died and not active. If a Patients with liver hydatid cyst preoperatively presenting with jaundice or biliary tree obstruction need investigated with magnetic resonance cholangiopancreatography (MRCP) (Fig 2)or endoscopic retrograde cholangiopancreatography (ERCP) for diagnosis or treatment. The choice of operative approach depended on the location of the cyst and the preference of the surgeon experiencess. After laparotomy or thoracotomy, the operative field was isolated by gauge packs soaked with scolicial agent as 10% hypertonic solution for prevention any spillagecyst fluid in to the peritoneal cavity during manipulation and evacuation During aspiration if cyst contents was bile-stained or purulent, the cyst classified as occult of complicated cysts.After evacuation of cysts, The cavity bottom was carefully examined for site of bile leakage and should be sutured. For obliteration of the residual cavity, surgical approach are either external drainage with folly catheter, capitonnage or omentoplasty.

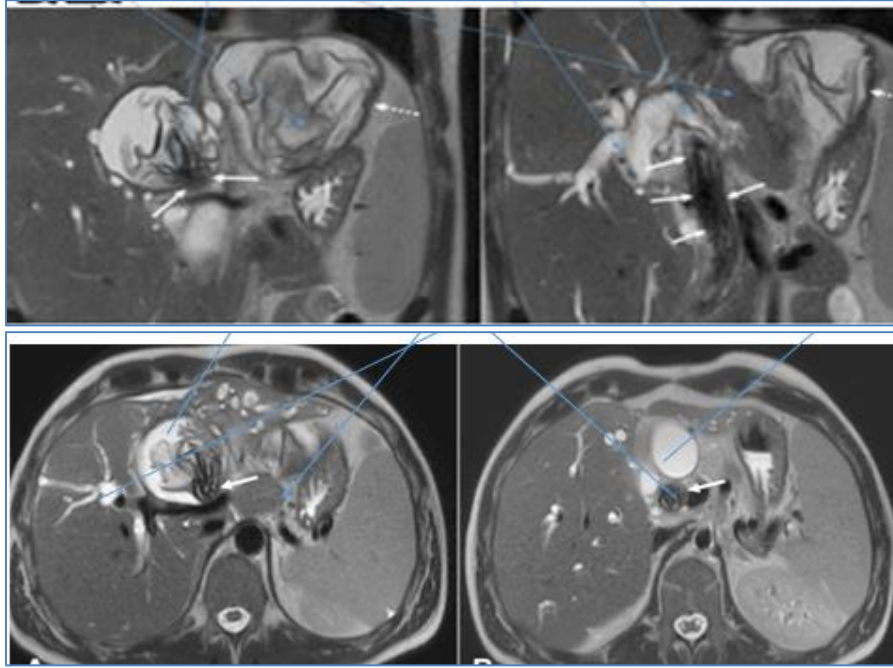


Figure 2 MRCP showing communication of the hydatid cyst with the biliary tree

Medical treatment with Albendazole for prophylaxis was administered one week preoperative (10 mg/kg/day). And continue postoperatively in three cycle of 28-day followed by a 14-day free interval. Liver function tests were scheduled regularly. Because hepatitis present in some patients. Data was collect and analysed.

3. Results

Fifteen patients with intraabdominal rupture, Ten of the patients were men and five were women. Mean age was 31 years. All of the patients had severe to mild tenderness, guarding, two shoulder pain and four mild to moderate allergic reaction. Three patients had a history of blunt abdominal trauma, two patients were during treatment with Albendazole. Six patients had fever, 11 had elevated white blood cell count, three patients had increased amylase (350 to 460) and four patients with elevation of ALT, AST, Bilirubin and Alkaline phosphatase. Ultrasound (US) and Computed tomography scans (CT) were obtained for all of the patients. (US) and (CT) revealed intra-abdominal fluid in all cases, intraperitoneal multiple elements and debris of cysts in 9 cases. (Fig 3). Moderate dilatation of biliary ducts due to intrabiliary rupture were seen in three cases (4, 5). All patients underwent surgery within the first 48 hours after presentation. One to three liters of hydatid fluid with floating daughter cysts, debris and purulent material was present in the abdomen (Figure 7). Evacuation, partial pericystectomy, and drainage was the most frequent surgical procedure. In two patients, there was direct adhesion between the cyst and the gallbladder, and cholecystectomy was performed (Fig 4). Procedures to fill the cystic cavities were, capitonnage and external drainage with Foley catheter in ten patients, omentoplasty in five patients. No patients died in hospital and postoperative period. A total of one or three complications developed in eight patients. biliary fistulae in three, wound infection in two and atelectasis in three patients. Median hospital stay was 9 days (range: 6–16 days) and median follow-up was 12 months (1–36 months). No recurrence developed in patients.

Frank intrabiliary ruptured hydatid cysts (IBR) of the liver were determined in 14 of the patients and occult IBR was determined in 32 patients. The average age of patients was 28 to 64 years. The most frequent symptoms were right upper quadrant, jaundice, fever and pruritus. Diagnosis of hydatid cyst was principally made using (US), (CT), MRCP and all patients underwent operation and choledochotomy, evacuation and lavage of the CBD was performed and remnant cavity irrigated with saline 7%. In frank IBR, during operation we found dilated CBD and daughter cysts, laminated membrane and debris in the CBD in all 14 patients but in the occult IBR needle aspiration during operation showed billows fluids (Fig 5). CBD irrigated with saline normal and evacuated all debris, daughter cysts and laminated membrane. Partial cystectomy and capitonnage were performed in 8 patients, omentoplasty in 6 patients plus T-tube drainage in all patients. In occult IBR, partial pericystectomy and capitonnage were performed in 22 patients, omentoplasty in 10 patients (Fig 1). In addition in 12 patients opening of the biliary fistula in the cavity was closed. In all cases a Foley catheter was put in the cavity. The most complication was pulmonary in 4, wound infection in 3 and biliary fistulae in 3 patients. A high output more than 400 ml biliary fistula occurred in one patient 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. The average period of hospitalisation was 16 to 28 days (range 8 to 12 days). T-tube drainage was removed after T-tube cholangiography (Fig 6).

15 patients with infection, Ten of the patients were men and five were women. Mean age was 31 years (range: 22 -66 years). All of the patients had signs of peritoneal irritation. Six patients had fever, 11 had elevated white blood cell count. (US) and (CT) were obtained for all of the patients and show an air fluid level inside the cyst (Fig 7), while the size of the cyst remains intact. Treatment was laparotomy, aspiration of pus, evacuation, pricstectomy and external drainage. Outcome was good. Site of cysts, Surgical procedures, and postoperative morbidity was shown in (Tab 1, 2, 3) In this study Albendazole was used at least 3 month postoperatively (10 mg / kg/ daily

Ten patients had pleural and pulmonary parenchymal complications 7 male and 3 female age ranging from 4 to 64 years. Pleural complications in 7 patients. The most Pleural complication was empyema and pleural effusion in 4 patients, hydropneumothorax in three patients and hepatobronchial fistula in 3 patients (Fig 7). The most common symptoms were chest pain and dyspnea. The most common finding in radiologic investigations were air fluid level. The most common procedure was Posterolateral thoracotomy with pherotomy in all patients, cystotomy, evacuation and decortication in 4 patients with omentoplasty or capitonnage and segmentectomy resection was performed in 3 patients with hepatic bronchial fistulae. One patient with intera pericardial rupture underwent anterolateral thoracotomy and pricardiotmy and evacuation all element of cyst and phrinotomy and evacuation of liver cyst and capitnnage. The mean postoperative hospital stay was 14 days (6-25 days). One patient died. Postoperative complications was pneumonia, atelectasis, wound infection and persistent air leakage, others was shown. No recurrence was observed in our patients during follow- up period (13, 14). Site of cysts, surgical approaches and complications in this study shown in Table 1, 2, 3.

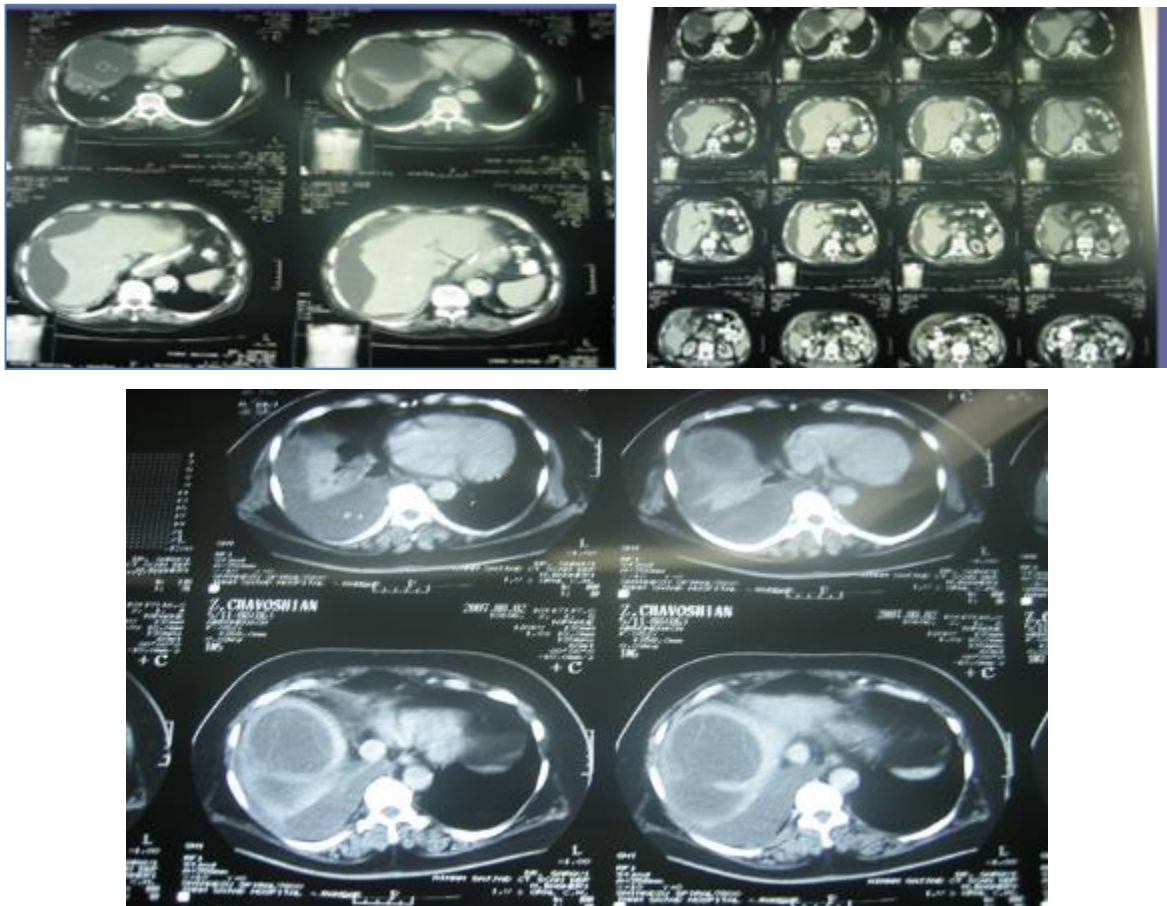


Figure 3 Interpritoneal cavity rupture

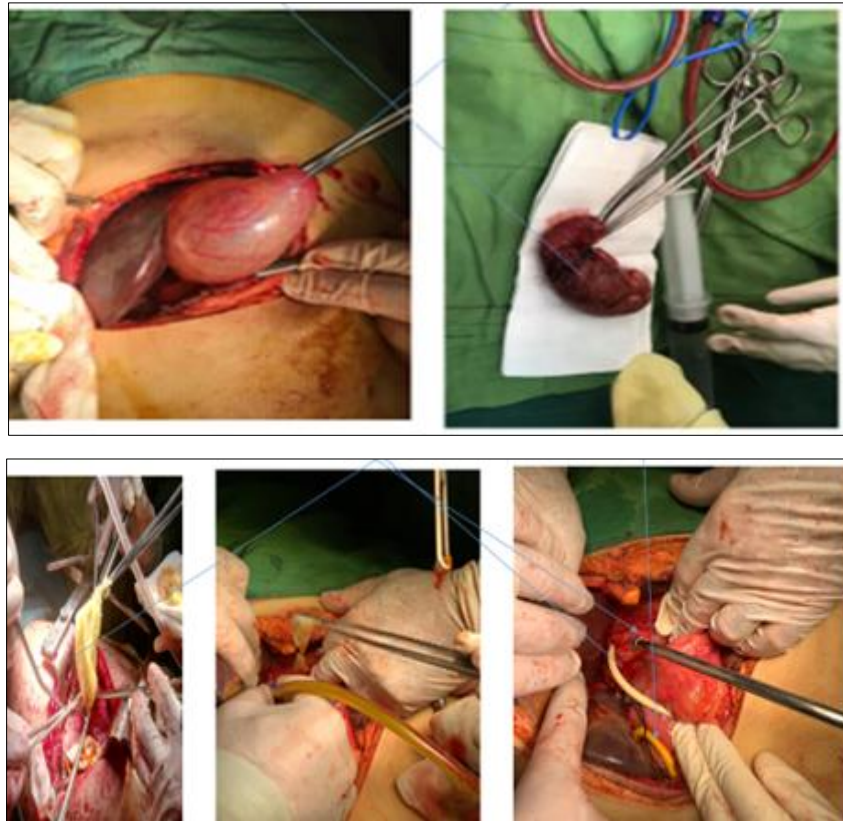


Figure 4 Cholecystectomy and T-tube insertion

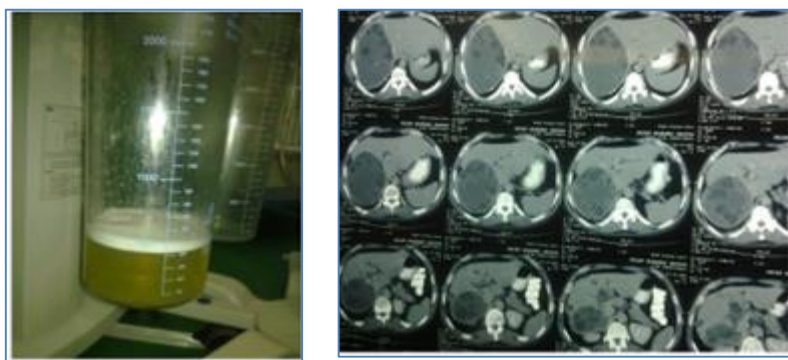


Figure 5 Occult cyst rupture whit bile suction fluid



Figure 6 Intraoperative and post operative cholangiography

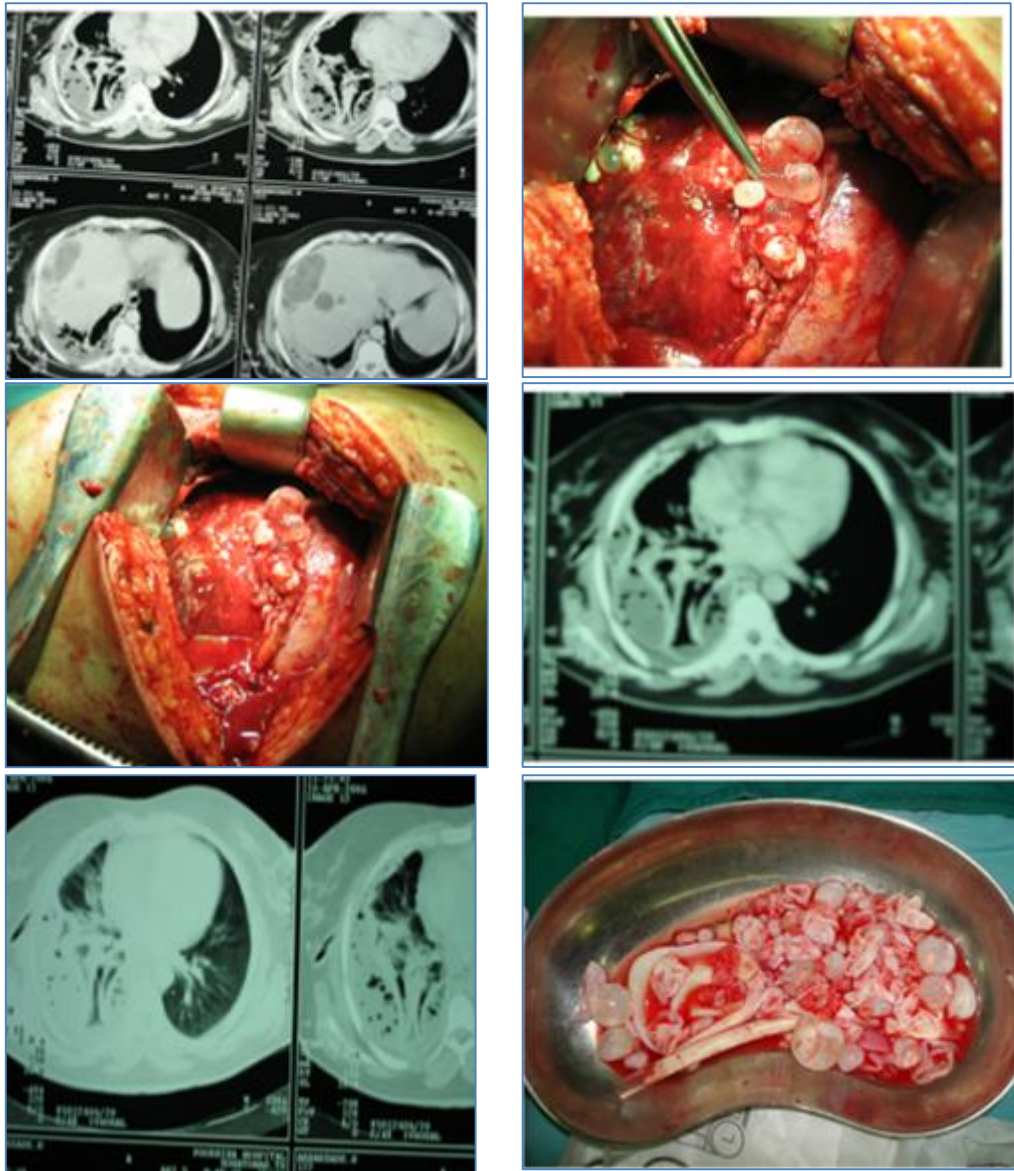


Figure 7 Rupture of hydatid cyst of liver in the pleural space .after thoracotomy and decortication and multiple daughter cysts present the view of air bubble

Table 1 Site of liver hydatid cysts

Right liver lobe only 10
left liver lobe only 5
both liver lobes only 4

Table 2 Methods of Surgery in 86 patients

Partial peri cystectomy + external drainage
Total Peri cystectomy + external drainage
cap tonnage + Peri cystectomy + external drainage
omentoplasty + drainage =5
capitonnage + Pericystectomy + external drainage
omentoplasty + external drainage =5
omentoplasty + external drainage +T-tube
ERCP +omentoplasty + external drainage +T-tube

Table 3 Postoperative Morbidity in 86 patients

Total complications =22
Cavitary abscess= 4
Biliary fistula = 6
Prolonged ileus = 4
pulmonary infection =5
wound infection=6
Plural empyema=4
Pleural effusion=4
•In a patient may more than one complication was occurred

4. Discussion

Complication of liver hydatid cysts (CLHC) are not uncommon. The incidence of (CLHC) reported in papers ranges between 30% to as high as 60% of patients diagnosed and treated for liver hydatid (5, 6–8,). In the present series, (22.36%) of the patients treated for hydatid cysts had CLHC). The (12.42%) of complicated cases were detected preoperatively (frank rupture) and (9.97%) intraoperative (occult rupture) show a lower rate compared with other series (30.3%–34.7%) (5, 9, 10, 11, 12, 13). The most common complications in our study was intera abdominal rupture second was intrabiliary rupture and third was suppuration of the cyst, which is also a different finding in literature (5, 14, 15, 16). Percutaneous aspiration injection and respiration (PAIR) and video laparoscopy are minimally invasive techniques which have been used for treatment (5, 17). We don't use these procedures. But today's Open surgery is the treatment of choice by the surgeons in this study (5). Recently, many surgeons recommended total pericystectomy and liver resections as the ideal treatment for liver hydatid cystic because they demonstrate lower recurrence rates (8, 5, 17). These more radical surgical methods have become available and safe but we never do this radical resection, because complication is high and no influence on the recurrences (19, 20). As a result, conservative surgical were more frequently used by our team (1, 3, 4, 12, 20). Cyst location and type of complications, and surgeons preference also influenced the choice of operative method (21). incidence of postoperative complications in the CLHC group are higher but the selection of best method may decrease the rate of and complications (1, 2, 3, 5, 8, 20). Supplementary postoperative Albendazole is indicated in uncomplicated and complicated cases, rupture of the cyst into adjacent structures or the peritoneal or pleural cavity and should be disseminated the element of cysts and disease (5, 8, 10, 14, 18, 20).

Intrabiliary rupture of hepatic hydatid cysts represents the most frequent complication of liver hydatid cysts having an incidence rate of 1%–25% in some reports (1, 5, 10, 11, 22, 23). According to our results, 14 of the patients were either diagnosed preoperatively (frank IBR) or 32 patients, bile-stained cyst content was found intraoperatively (occult IBR). Cystobiliary communication results from compression of bile ducts from the gradually increasing cysts, causing bile

stasis(1, 14, 5, 24).. The majority of the patients with intrabiliary rupture in our series (9.97%) did not manifest jaundice on initial presentation or preoperatively. Silent rupture involves small bile ducts, while symptomatic rupture is associated mainly with larger ducts and probably extrahepatic bile tree(5, 22). Biliary colic, cholangitis, jaundice, and more rarely, the presence of germinal membranes in feces are the expected manifestations(5, 22, 26).

The management of IBR depended on the preoperative and intraoperative findings(5, 14, 17, 19, 22). In our study laparotomy, evacuation, partial pericystectomy and search for the orifice of the possible fistula was the first attempted and close it with stiches.And in next step with A clean gauze pad with hypertonic saline was applied inside the cyst and continue for 10 min in order to find the orifice. In some cases, the orifice of the fistula not present and no more management needed. After evacuation and pricystectomy, Choledochotomy needed and with a catheter irrigation of the common bile duct from hydatid cysts debris and daughter cysts and piece of laminated membrane needed. placement a T-tube, Intraoperative cholangiography used in order to search for parasitic material inside biliary tree or to detect the cystobiliary communication(1, 14, 22, 27). Cholecystectomy was performed in two patients because of sever adhesion between gallbladder and cysts. indications of cholecystectomy are the presence of a fistula between the hydatid cyst and the gallbladder and sever adhesion between cyst and gallbladder and the primary hydatidosis of the gallbladder (28, 29, 30).

ERCP is a valuable method for management of frank IBR, MRCP is also used for diagnostic IBR (1, 27, 31, 32, 33). when clinical symptoms of acute cholangitis are present ERCP is indicated and endoscopic papillotomy should be performed when hydatid cyst debris is detected inside the common bile duct (1, 5, 31, 32, 33). when postoperative the presence of a biliary fistula with persistent high output, remaining hydatid material in the common bile duct, and jaundice, endoscopic papillotomy indicated (31, 32, 33). Vignote et al. (33) reported that persisting fistulas usually close 5–7 days after papillotomy. In our series, 2patients underwent endoscopic papillotomy postoperatively and stopped leakage.

Infection of the hydatid cysts was 23% of cysts managed in some series (34, 5). The presence of cyst leakage is essential for bacterial contamination of the endocystic fluid, and therefore, it is believed that cystobiliary communication is the more frequent cause of infection (34, 35, 26). But in our study the infection rate was (4.67%). Clinical presentations are fever, chills, sepsis and abdominal pain as in liver pyogenic abscess.

Intera abdominal rupture of hydatid cysts is a more serious complication. Abdominal trauma in a patient with large, superficial, thin-walled cysts in the anterior and inferior part of liver are high risk for cyst rupture (1, 5, 13, 14, 15, 27), although spontaneous rupture is reported (4, 7, 10, 15). Clinical presentation includes anaphylactic shock, acut abdomen, pruritus. (1, 3, 13, 14, 15,). Surgical management is emergency aims a are to removal of all the cysts material and profuse peritoneal irrigation with scolicidal agents betadine for prevention of recurrence (1, 5, 13, 22, 26).

Cysts located at dome of the liver surface may perforated the diaphragm and open in to the pleural cavity and lungs parenchyma (5, 12, 21). The incidence of perforation of diaphragmatic and extend to cavity of the ranges from 0.6% to 16% of cases with hepatic echinococcosis (5, 12, 36, 37). in our series was 3%. Erosion of Diaphragm lead either or penetration of lung parenchyma resulting in pneumonitis or lung abscess (5, 12, 22, 36, 37). In the case of hydatid cyst ruptures to the lumen of a bronchiole, daughter cysts may appear in the sputum (5) one of our patients' expectorations of bile secretion or daughter cysts. a Broncho biliary fistula is presented. Surgical treatment in bronchial fistula is suturing of the fistulae, Wedgeresection, segmental resection, or lobectomy, management of the liver cyst and repair of diaphragm, decortication of pleural space,, and external drainage of liver cyst remnant cavity Chest –tube(5, 12, 36, 37).

When a thoracic empyema or effusion present transthoracic approach is more perfect, when exploration of common bile duct is necessary laparotomy is selected (5, 12, 21, 22, 36, 37,).

5. Conclusion

Complicated hydatid cyst of the liver can be successfully managed surgically with good long-term results. Complicated liver echinococcosis demonstrates several distinct features that differentiate it from the noncomplicated disease. Frequently severe clinical manifestations, complexity of surgical management, and the increased postoperative complications characterize complicated liver echinococcal disease in the Intrabiliary Every effort should be made to detect and prevent these complications, ERCP and MRCP can helpfor diagnosis and treatment. With appropriate evaluation and intervention, this will lead to decreased postoperative bile leakage and fistula formation, a major cause of morbidity and mortality.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of ethical approval

This case Report Approved by Teaching and Ethic comity of ARYA Hospital.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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