ORIGINAL ARTICLE

Surgical management of obstructive jaundice due to spontaneous intrabiliary rupture of hydatid cysts of the liver

Sheikh Atiq-ur-Rehman, Muhammad Siddique, Ahmad Yar, Gulzar Ahmad, Muhammad Mazhar Bashir

Received:
6th July 2016

Accepted:
9th October 2016

Quaid-e-Azam Medical College& Bahawal Victoria Hospital (QAMC/BVH)
SAur-Rehman
M Siddique
A Yar
G Ahmad
MM Bashir

Correspondence:
Dr Sheikh Atiq-ur-Rehman, House No: 9/A, Block-X, Model Town C, Bahawalpur
Cell:0300-6808500
mail:dratiq2002@gmail.com

Abstract

Purpose: To evaluate the clinical presentation, operative findings & postoperative course of patients with intrabiliary rupture of hydatid cyst of the liver.

Place & period: The study was conducted in surgical units, Bahawal Vicotria Hospital, Bahawalpur, Pakistan from 01-01-2004 to 31-12-2015.

Material & method: This was a prospective type of observational case series study. All patients of either sex & age being diagnosed as case of obstructive jaundice due intrabiliary rupture of hepatic hydatid disease were included in the study. Using a standardized data collection instrument, clinical case record of all patients who were operated for intrabiliary rupture of hydatid cyst of the liver was noted.

Results: 20 patients (13 males, 7 females) with intrabiliary rupture of hepatic hydatid cyst were enrolled. The age range was from 30-70 years. The most frequent symptoms were right upper quadrant/epigastric pain, jaundice and pruritus. Diagnosis of hydatid cyst was principally made using ultrasonography. 14(70%) cysts were located in the right lobe, 3 (15%) in the left lobe, and 3 (15%) in the both lobes. The size of the cysts was from 05-20 cm. Common bile duct (CBD) was dilated in 40% patients. CBD exploration was done in all patients. T-tube drainage of CBD was done in 18(90%) patients. Choledochoduodenostomy was done in 2(10%) patients. Postoperative course was uneventful in 12(60%) patients. Wound infections developed in 3(15%) patients, suppuration of the residual cavity in 3(15%) patients, septicemia and renal failure in 2 (10%) patients and wound dehiscence & GI bleeding in one patient each. The hospital stay was 7-25 days. All patients were given 8 week course of albendazole or mebendazole postoperatively. Two patients (10%) died from sepsis & multiple organ failure.

Conclusion: An unusual cause of obstructive jaundice due intrabiliary of hepatic hydatid cyst is daughter cysts and or sludge in the CBD. If a dilated CBD and bile-stained cystic fluid is found in patients with hydatid disease of the liver, common bile duct exploration should be performed during operation. Surgical removal of the cyst from CBD resulted in complete resolution of jaundice. Treatment with oral albendazole was given pre-operatively and continued for two months after surgery.

Keywords: obstructive jaundice, hepatic hydatid disease, echinococcus granulosus

Introduction:

Hydatid cyst is caused by Echinococcus granulosus. The humans are the accidental Intermediate host.[1] The most common sites are the liver account for 60%, the lung account for 30% although it may develop at other sites including kidney, bone, brain and heart.[2] Hepatic hydatid disease (HHD) is a major endemic problem in sheep-rearing regions of the world.[1,3] The liver acts as a filter for hydatid larvae, making it the most commonly affected organ.[4,5] Up to one-third of patients with HHD present with complications such as rupture (into the biliary tree, thorax or peritoneum), secondary infection, anaphylactic shock, sepsis and liver replacement.[7,12,13]

Intrabiliary rupture is the most common complication of hepatic hydatid cyst yet it is unusual, occurring in only 3–17% of cases.[3,4,12,13] Communication of a hydatid disease with the biliary tree has been described in up to 90% of hepatic hydatid cysts.[5] This is due to the incorporation of biliary radicles into the pericyst. However, frank rupture into the biliary tree occurs in only 5–15% of cases.[3,12] Although the communication with the intrahepatic bile ducts is common, its occurrence between a hepatic hydatid cyst and the gallbladder is rare.[5,6,17] It presents clinically as pain, obstructive jaundice, cholangitis or sepsis, and can be fatal without intervention. [1,2,7,8,9] Successful outcome depends on accurate pre-operative diagnosis. Diagnosis of this complication can usually be made using ultrasound and CT scan.[7, 8,10]

On imaging studies, a dilated intrahepatic bile duct segment in association with an adjacent cystic lesion and dilatation of the common bile duct are generally the indirect findings of rupture of a hydatid cyst into the biliary tree. Ultrasonography may identify the daughter cysts within the biliary tree. The fistulous communication between the hydatid cyst and biliary tract may sometimes be directly identified on different imaging studies.[7,8,10] Complete evacuation of the contents of a liver hydatid cyst is very rare and can present diagnostic problems.[6]

MRI and MRCP are used in difficult cases in which CT scan and ultrasonography findings may be inconclusive. The wall of the hydatid cyst is seen as a low intensity rim, a reliable sign to differentiate hydatid cyst from other simple cysts. Daughter cysts have a lower signal intensity compared with the mother cyst. The MRI finding in ruptured hydatid cyst can be direct or indirect. A breach in the low intensity rim of the cyst wall with extrusion of cyst contents is a direct sign, while increased echogenicity, fluid levels, presence of air and changes in signal intensity are indirect signs.[3,7,11]

ERCP is the gold standard with which to confirm biliary tract involvement and may be of therapeutic value in selected cases. On ERCP, a swollen ampulla of Vater may be seen, with hydatid material protruding out. Dilated ducts with debris and daughter cysts may appear as radiolucent filling defects. Irregular leaf-like material that changes shape with changes in pressure differentiates this condition from other causes of obstructive jaundice.[13,15] HIDA can be helpful in doubtful cases with cystobiliary communication where ultrasound and CT are not conclusive.[11]

The chemotherapy has been disappointing results & for this reason surgery remains the mainstay of treatment for hydatid cyst rupturing in to biliary system. The accepted treatment for intrabiliary rupture of hydatid cyst is surgical decompression of CBD and removal cyst from the liver.[16,19,20]

Patients & Methods:

It was a prospective observational case series study which was conducted in surgical units, Bahawal Victoria Hospital Bahawalpur, Pakistan from 01-01-2004 to 31-12-2015. Total twenty patients were presented with obstructive jaundice due Intrabiliary rupture hepatic hydatid disease(HHD) during the study period. Patients of any age and both gender, who presented with symptoms & signs obstructive jaundice and diagnosed as a case of Intrabiliary rupture of hepatic hydatid cyst was the cause were included in the study. All those patients who have obstructive jaundice due other causes for example CBD stone, stricture or tumour were excluded. These patients of obstructive jaundice were presented to surgical OPD as well as to emergency department and also referred to surgical ward from medical ward. The clinical record of the patients was made on a performa. It has a record of patient's biodata, symptoms & signs, biochemical & radiological report, treatment (surgery) given and a follow-up. The diagnosis was made by using ultrasound and CT scan. All patients of intrabiliary rupture of HHD were operated. Before commencing for the operation hemoglobin, glucose, fluid and electrolyte were corrected. A broad spectrum antibiotic and vitamin K was also given. Postoperative care included nil by mouth, naso-gastric aspiration and intra-

Table 1: Clinical presentations of intrabiliary rupture of hepatic hydatid disease. Total no of patients 20.

Clinical presentation	No of patients (%)
Jaudice	20 (100%)
Pain in RHC	08 (40%)
Itching	06 (30%)
history of cholecystectomy	02 (10%)
Fever, weight loss, anorexia, nausea/vomitting	03 (15%)
Mass right hypochondrium	05 (25%)
Hepatomegaly	08 (40%)

Table 2: Results liver function test. Total no of patients 20.

No	serum bilirubin (mg/dl)	SGPT Level	Alkaline Phosphatase	No of patients
1	3.8 - 5.9	31-42	380-470	10(50%)
2	6.2-9.0	23-45	485-677	5(25%)
3	10.1-13.8	28-55	690-778	3(15%)
4	14.0-15.8	30-85	792-887	2(10%)

Table 3: Results of ultrasonography. Total no of patients 20.

No	condition	No of patients
1	Cysts size 05-10 cm	11(55%)
2	Cysts size 10-20 cm	09(45%)
3	CBD dilatation 08-10mm	12(60%)
4	CBD dilatation 12-15 mm	06(30%)
5	CBD dilatation 17-20 mm	02(10%)
6	Gall stone	02(10%)
7	Right lobe of liver involved	14(70%)
8	Left lobe of liver involved	03(15%)
9	Both lobes of liver involved	03(15%)

Table 4: Hospital stay. Total no of patients 20.

No	Hospital stay in days	No of patients	%
1	07-09	02	10
2	10-12	06	30
3	13-15	08w	40
4	16-25	04	20

venous fluid, vitamin K and broad spectrum antibiotic. T-tube cholangiogram was done in patients where it was used for drainage of CBD on 7th post-operative day. Computer programmed SPSS was used for analysis.

Results:

In our study, 20 cases of obstructive jaundice due to intrabiliary rupture of hepatic hydatid disease presented to surgical units of Bahawal Victoria hospital Bahawalpur, Pakistan. These patients presented to surgical units from 01-01-2004 to 31-12-2013. Out of 20 patients, 13(65%) were male and 7(35%) were female. The age range was 30-70 years. The majority of patients presented in fourth decade of their life. 75% patients were resident of rural area of Bahawalpur and 25% patients from urban area. 80% patients in our study belonged to low socio-economic class of the society.

Patients with intrabiliary rupture of hydatid cyst of liver leading to obstructive jaundice have a variable clinical course. All patients have jaundice at the time of presentation to surgical unit. Symptoms & signs were shown in table no: 1. Haematological and biochemical profile especially liver function tests e.g. serum billirubin, serum transaminase and alkaline phosphatase level were done in all patients. Results of liver function tests were given in table no: 2. The ultrasonography has high accuracy rate but it is operator dependent. On ultrasonography hydatid cyst in liver was found in 100% patients. Hydatid cyst in right lobe of liver in 14 (70%) patients and in left lobe of liver in only 3(15%) patients. 3(15%) patients had hydatid cyst in both lobes of liver. The cyst size ranges from 5-20 cm, showing variable ultrasound findings likes cystic lesion, fluid collection with cart wheel appearance, fluid collection with split wall sign and mixed hetrogenous echogenicity pattern. Gallstone was present in two patients. Common bile duct was dilated in 40% patients, in 12(60%) patients CBD size was 08-10mm, in 6(30%) patients CBD size was 12-15mm and in 2(10%) patients CBD size was 17-20mm (table no: 3). Eleven patients had history of drug treatment (mebandazole, albendazole) for variable period. No previous history of drug treatment was found in 9 (45%) patients. All patients were operated; pre and per-operative assessment was done to select the surgical procedure performed in each patient. On exploration of CBD, daughter cysts and debris found in 16(80%) patients, T-tube drainage of CBD in 18(90%) patients and choledochoduodenostomy in 2(10%) patients performed. Hepatic hydatid cyst was incised and daughter cysts were removed carefully after isolating the area with hypertonic saline

swab in all patients. An internal opening of the biliary fistula was found and sutured in 12(60%) patients. Partial cystectomy/ cystectomy and capitonage or omentoplasty or drainage or marsupialization was performed in all patients. 12(60%) patients had uneventful recovery. Post-operative complication like wound infection in 3(15%) patients, suppuration of the residual cavity in 3(15%) patients, septicemia and renal failure in 2(10%) patients. One patient developed gastro-intestinal bleeding and another patient developed wound dehiscence. 2(10%) patients died from sepsis & multiple organ failure. All patients were given 8 week course of albendazole or mebendazole postoperatively. The hospital stay was 7-25 days (table no: 4). The hospital stay was long for patients who developed septicemia, renal failure and had associated systemic medical problem for example hypertension, ischemic heart disease or diabetes mellitis. All the patients were advised follow up for at least three months but only 50% patients followed the advice. At the time of visit, clinical examination of patients was carried thoroughly; liver function tests and ultrasonography for liver & biliary system were done.

Discussion:

Hepatic hydatid disease (HHD) is the commonest form of parasitic infestation caused by Echinococcus granulosus. It is an endemic problem in sheep-rearing regions of the world.[1] Of all cysts in the liver, 50%-70% are caused by Echinococcus granulosus. The right lobe of the liver is affected in 80% of cases and the left lobe in 20%. HHD may be asymptomatic, or may cause abdominal pain, jaundice, or a visible abdominal mass.[1,2] Patients seek medical care when their cysts have reached large size destroying a large amount of liver parenchyma. In our study most of the patients (75%) who were admitted in surgical unit of B-V hospital Bahawalpur with obstructive jaundice due to HHD came from rural areas. 80% patients in our study belonged to low socio-economic class of the society.

There are many potential local complications of HHD such as intrahepatic complications, exophytic growth, transdiaphragmatic thoracic involvement, perforation into hollow viscera, peritoneal seeding, portal vein involvement, abdominal wall invasion, and frank biliary communication (i.e intrabiliary rupture of the cyst) which has been reported in only 5–15% of cases.[1,2,3] Rupture into the biliary tree is although uncommon complication of HHD. It occurs into the right duct in 55–60% of cases, into the left duct in 25–30% and rarely into the confluence or gall bladder.[5,6,8,17] Lewall and McCorkell have classified rupture of hydatid cysts into three types: contained, communicating and direct. Communicating rupture can be simple communication between small biliary radicles and the cyst, or frank rupture into the biliary tree.[9] When rupture into the biliary tract occurs, the cystic fluid escapes into the biliary tract with daughter cysts discharged into the common bile duct, causing biliary colic, obstructive jaundice, and possibly liver abscess fever, acute cholangitis, abdominal lump, allergic manifestations and rarely with acute pancreatitis, or septicaemia, or it may be asymptomatic (5–6%).[3-5,12-14] Clinical presentation of patients was variable in our study. Jaundice was present in 100% patients in our study. Pain right hypochondrium with was present in 40% patients and mass right hypochondrium was pesent in 25% patients. Acute cholangitis was present in 15% of patients in our study. No patient in our study presented with acute pancreatitis or allergic manifestation of intrabiliary rypture of HHD.

A correct diagnosis of intrabiliary rupture using ultrasound was possible in 66.6–94% of cases. The presence of a questionable hepatic focus is generally a prerequisite for a radiologist to even consider the differential of intrabiliary hydatid rupture in a patient having echogenic material in the biliary tree. Ultrasonography showed a hydatid cyst in the liver (heterogeneous echogenic interior and a ring like pattern of calcification), dilatation of the extrahepatic biliary radicals, echogenic or non-echogenic material without posterior acoustic shadowing is seen in the biliary tree, suggestive of sludge and daughter cysts, or visualization of the communication between the cyst and the biliary tree. In a frank communicating rupture, the cyst becomes smaller, and

undulating membranes may be seen within it.[7,10] On ultrasonography hydatid cysts was found in liver in all patients but dilated CBD was present in 40% patients in our study. Hydatid cyst was in right lobe of liver in 14 (70%) patients, in the left lobe of liver in 03 (15%) patients while 03 (15%) patients had hydatid cyst in both lobes of liver. In 12(60%) patients size of CBD was between 08-10mm, in 6 (30%) patients size of CBD was between 12-15mm and in 2 (10%) patients size of CBD was between 17-20mm. These features in our study were similar to the literature.

The features of a hydatid cyst on CT scan are enhancement of the cyst wall and the internal septae, visualization of detached undulating membranes and calcification of the cyst wall. A dilated CBD with low attenuation intraluminal material suggests the presence of hydatid sand and cysts in the CBD. An interrupted area of the cyst wall proximal to a dilated duct may be identified as representing the site of communication. Cyst wall discontinuity, a direct sign of rupture, was seen in only 75% of cases. The accuracy of CT combined with ultrasound was near 100% in cases with uncomplicated intrabiliary rupture.[7,8,10] In our study diagnosis of obstructive jaundice due intrabiliary rupture of HHD was made on USG because hydatid cyst in liver was present in all patients.

Ultrasonography and CT scan are the most widely used modality for detecting intrabiliary rupture of HHD,[7,10] but rupture of the cyst and evacuation of its contents may change the radiological appearance necessitating more sophisticated investigations such as MRI, MRCP, endoscopic retrograde cholangiopancreatography (ERCP) and hepatobiliary scintigraphy.[8,11,15]

Jaundice due to extra-hepatic biliary obstruction may be caused by many diseases some of which are quite rare.[2,19] In all cases of surgical jaundice, it is mandatory to determine pre-operatively the exact nature and the site of obstruction because an ill-chosen therapeutic approach can be dangerous. Operative decompression may relieve extra-hepatic blockage but operation can only harm patients with intra-hepatic block, parenchymal cell inflammation or necrosis.[8,13,14] Pre and per-operative assessment was done to select the surgical procedure performed in each patient. All patients in our study were operated. Hepatic hydatid cyst was incised and daughter cysts were removed carefully after isolating the area with hypertonic saline swab in all patients. Partial cystectomy and capitonage or omentoplasty or drainage or marsupialization was performed for hydatid cyst in liver.[16,19] An internal opening of the biliary fistula was found and sutured in 12(60%) patients in our study.

The classical treatment of HHD ruptured in to the biliary tract was exploration of common bile duct i.e choledochotomy. Then clearance of daughter cyst & sludge from CBD and placement of T-tube in CBD is best to decompress the biliary tree or choledochoduodenostomy.[16,18,20] In our study on exploration of CBD, daughter cysts and debris found in 16(80%) patients, T-tube drainage of CBD in 18(90%) patients and choledochoduodenostomy in 2(10%) patients performed. Postoperative course was uneventful in 12(60%) patients. Postoperative complication like wound infection & suppuration of the residual cavity in 3(15%) patients, septicemia and renal failure in 2(10%) patients. One patient developed gastro-intestinal bleeding and another Wound dehiscence. Two patients (10%) died from sepsis-multiple organ failure and hepatic failure.

Conclusions:

Intrabiliary rupture of hepatic hydatid cyst is rare, but important cause of obstructive jaundice. Surgeons should be aware of this complication of HHD even where the disease is not endemic. The cause of obstructive jaundice due intrabiliary of HHD is daughter cysts and or sludge in the CBD. The diagnostic investigations include abdominal ultrasonography and CT scan. If the diagnosis is confirmed, surgery and adjuvant chemotherapy is indicated. The classical treatment of hydatid disease ruptured in to the biliary tract was exploration of common bile duct (CBD) i.e choledochotomy and decompression i.e T-tube drainage/choledochoduodenostomy.

If the diagnosis is not confirmed preoperatively but a dilated CBD and bile-stained cystic fluid is found in patients with HHD, CBD exploration should be performed during operation.

Conflict of Interest: None

Funding Sources: None

Role and contribution of authors:

Dr Sheikh Atiq-ur-Rehman, FCPS(Surgery), MRCS(Eng), Assistant Professor Surgery, QAMC/BVH, Bahawalpur did the initial write-up and collected the data.

Dr Muhammad Siddique, FCPS(Surgery), FCPS(Paeds Surgery), Senior Registrar Paeds Surgery, QAMC/BVH, Bahawalpur helped in collected the data and collected the references

Dr Ahmad Yar, FCPS(Surgery), Senior Registrar Surgery, QAMC/BVH, Bahawalpur helped in collecting the references and writing of the discussion

Dr Gulzar Ahmad, FCPS(Surgery), Professor of Surgery, QAMC/BVH. Bahawalpur conceived the idea of writing the article and review the study and gave final touchup

Dr Muhammad Mazhar Bashir, P.G trainee surgery, BVH Bahawalpur helped in collecting data and also collecting references.

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