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Original Research Article

Cysto-biliary communication (CBC) in hepatic hydatidosis: predictors, management and outcome

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ABSTRACT

Background: Liver hydatidosis is a common health problem in endemic areas. Cystobiliary communication is the most common complication of liver hydatid.
Methods: Cases of liver hydatid operated during the period June 2012 to July 2018 were retrospectively assessed. Patients diagnosed with cystobiliary communication (preoperatively, intraoperatively or postoperatively) were analysed. Demographics, laboratory tests were noted. Computed tomographic (CT) findings including size, location, Gharbi's type, presence of intrahepatic biliary radical dilatation, CHD and CBD dilatation were noted. Intraoperative findings were noted. Postoperative outcomes and any intervention if done were noted. Results were analysed.
Results: Around 108 patients with liver hydatid underwent surgical intervention. Of which 20 (18.5%) patients were found to have cystobiliary communication. Mean cyst size was 8cms. Location of cyst in segment IV and V commonly. 8 patients were diagnosed preoperatively by elevated bilirubin and alkaline phosphatase and on contrast enhanced computerized tomography. Out of 8, 4 patients underwent preop ERCP and stenting, followed by surgery and the other 4 underwent direct surgery with CBD exploration. 4 were diagnosed intraoperatively and the fistula site sutured. But 2 patients had postop biliary fistula which required postop ERC and stenting. Remaining 8 presented postoperatively with biliary fistula. Of 8 patients, 2 had major and 6 had minor fistulas. Postoperative mortality was 0%.
Conclusions: Cystobiliary communication is more common in males with large cyst size, located in the central segments of liver close to biliary confluence, Gharbi type IV cysts. Timely diagnosis and appropriate management decrease the morbidity and mortality.

Keywords: Biliary fistula, Cystic echinococcosis, Cysto-biliary communication, Hydatid cyst

INTRODUCTION

Liver hydatid disease caused by *Echinococcus granulosus*, is a common public health problem all around the World.[1,2] Infestation of humans with larval form incidentally causes hydatid cyst formation in different organs, most commonly liver. Hepatic hydatid disease usually presents by abdominal pain or palpable lump. Rupture of the cyst is the most common complication as the cyst enlarges. The cyst may rupture into the peritoneal cavity, pleural cavity, blood stream or most commonly intrabiliary rupture.[3] Cystobiliary fistula is reported in 5%-42% of hepatic hydatid disease cases.[4] The cystobiliary communication (CBC) is classified into 2 main types: minor (simple or occult) fistula and major (frank) fistula. A minor fistula (10-37%) is usually asymptomatic and may be diagnosed intraoperatively or post operatively by an external biliary fistula.[5-7] Frank or

major fistula (5% to 17%) is a wide communication between the cyst and the biliary system that allows the contents of the cyst to drain into the biliary system causing obstructive jaundice, cholangitis, secondary infection of the cyst, or even anaphylaxis.[8] Cystobiliary communication increases the risk of perioperative morbidity and requires additional management, several studies were conducted to identify the predictors of CBC in hepatic hydatid disease. These predictors included cyst size >10cm, calcified wall, recurrent disease, preoperative jaundice, and preoperative elevated serum alkaline phosphatase (ALP).[9,10]

Aim of the research work was to study the presentation, risk factors and management of hydatid cyst with biliary communication.

METHODS

One hundred and eight cases of liver hydatid operated during the period June 2012 to July 2018 were retrospectively assessed at NRI General Hospital, Chinnakakani. This was the retrospective study. This study was conducted for 6 years. Recorded data was collected. Parameters analyzed are:

- Epidemiology,
- Clinical presentation,
- Risks factors assessed are total leucocyte count, alkaline phosphatase, serum bilirubin.

Inclusion criteria

All patients diagnosed with cystobiliary communication either preoperatively, intraoperatively or postoperatively were analysed.

Exclusion criteria

- Patients who underwent non surgical treatment,
- Patients with extra hepatic echinococcosis.

Procedure

This is a retrospective study. Recorded data was collected from the patient records. All the patients undergoing surgical treatment for Hepatic hydatidosis in the department of Surgical Gastroenterology during the study period i.e. June 2012 to July 2018 are selected and the patients with a cysto biliary communication were included in the study. Complete demographic, clinical, biochemical and radiological details of these patients were noted and analysed. The following parameters were analyzed as predictors of CBC-elevated serum bilirubin, elevated total leucocyte count and elevated alkaline phosphatise. The course of these patients, including pre-operative work up, intra-operative findings and procedure done for CBC, post-operative data were noted and analyzed. The final outcomes of these patients including post operative biliary fistula and their management were studied.

Statistical analysis

Descriptive and inferential statistical analysis has been carried out in the present study. Results on continuous measurements are presented on Mean±SD (Min-Max) and results on categorical measurements are presented in Number (%). Significance is assessed at 5 % level of significance. Chi-square/Fisher Exact test has been used to find the significance of study parameters on categorical scale between two or more groups.

RESULTS

Out of 108 cases of hydatid cyst of liver cases operated in our department during the study period 20 (18.5%) cases had cystobiliary communication (Figure 1). Mean age of presentation was 44.4years (45 to 52years) (Table 1). All the patients had pain abdomen on presentation, while 8 had jaundice on presentation. The total leucocyte count was more than 11000cells/mm^3 in 12 (60%) patients and less than 11000cells/mm^3 in 8 (40%) patients (Table 2).

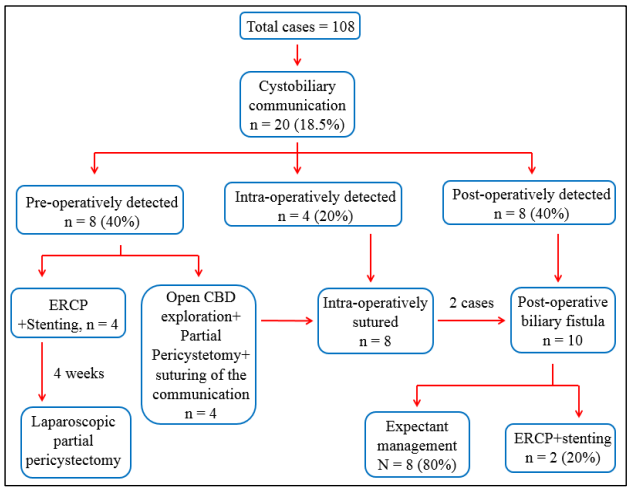

Figure 1: Study flow chart.

Table 1: Demographics.

Variable	Number (n)
Total no. of cases	108
Cases with cysto-biliary communication	20 (18.5%)
Mean age	44.4 years
M:F	1.5:1

The total bilirubin was elevated in 8 (40%) patients and it was normal in rest of the patients. Serum alkaline phosphatase was more than 120IU/L in 16 (80%) of the patients and it was less than 120IU/L in 4 (20%) of the cases. All patients underwent contrast enhanced computed tomography (CECT) (Figure 2) of the abdomen with liver protocol. The location of the cyst was

in segments IV and V in 70% of the patients. Gharbi's type was type IV in 12 cases and type II and III in other 8 cases. Intra hepatic biliary dilatation, common hepatic duct and common bile duct dilatation was noted in 10 (50%) patients. The mean cyst size was 8cms (6.4-10.8cms).

Table 2: Clinical and biochemical and radiological parameters.

Variable	Number (n)
Presenting complaint	
Pain abdomen	20
Jaundice	8
Biochemical parameters	
Total leucocyte count	
>11,000cells/cu.mm	12
<11,000cells/cu.mm	8
Serum total bilirubin	
>2.5mg/dl	8
<2.5mg/dl	12
Serum alkaline phosphatase	
>120IU	16
<120IU	4
Radiology parameters	
Mean cyst size	8.4cms
Gharbi type 4	12
Gharbi 2 & 3	8

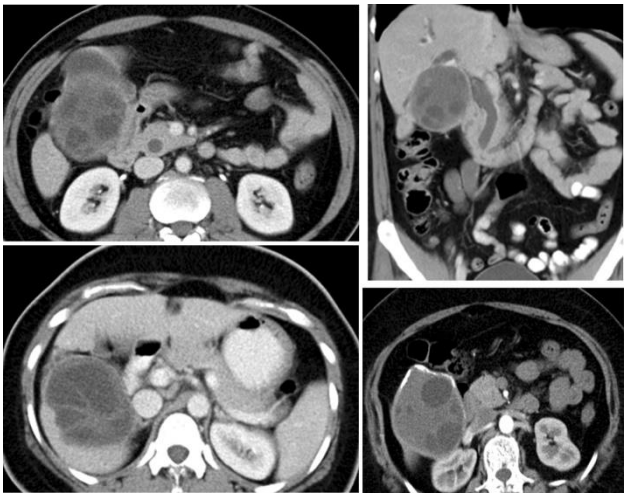

Figure 2: CECT images.

A communication was identified in the preoperative imaging in 8 cases. Four of these cases underwent preoperative Endoscopic Retrograde Cholamgiopancreaticography (ERCP) (Figure 3A) followed by laparoscopic pericystectomy after 4weeks. The remaining four patients underwent open common bile duct exploration with partial cystectomy (Figure 3B, 3C) with suturing of communication. A cystobiliary communication (Figure 3D) was identified intra-operatively in four patients during laparoscopic surgery and suturing of the communication was done. Two patients in his group developed post-operative biliary fistula which subsided with conservative treatment. In the remaining eight patients, no biliary communication was identified intra-operatively, but they developed biliary fistula post-operatively. In six patients, the fistula subsided with conservative management but two patients required ERCP with biliary stenting.

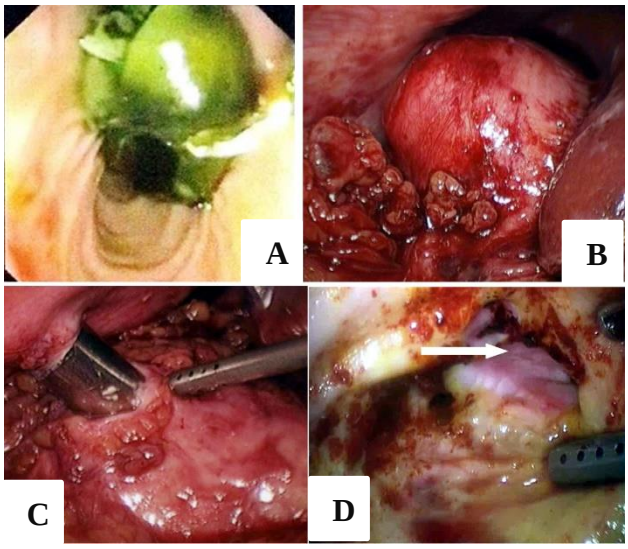

Figure 3: A) ERCP image showing extraction of membranes. B) Intra-operative image showing the cyst. C) Extraction of the contents by introducing port directly into the cyst. D) Cysto-biliary communication (CBC).

DISCUSSION

Hepatic hydatid disease (HHD) remains a medical problem of great concern. Intrabiliary rupture is the most common complication of hepatic hydatid cyst. Its incidence in literature ranges from 1.0% to 25.0%, in our series it was 18.5%.[3] cysto-biliary communication is a major risk factor for post-operative morbidity, pre-operative and intra-operative diagnosis and management helps to effectively enhance the post-operative outcome. Predicting the presence of CBC begins with pre-operative factors detected in the history, examination, laboratory, and imaging findings. Obstructive jaundice and/or cholangitis are sensitive indicators of the presence of a frank CBC in more than 60% of the cases.[11] An intracystic pressure around 35cm H_2O is an indicator of its viability, and this pressure is generally higher than the intraluminal pressure of the biliary radicals. This high pressure is the cause of CBC. Two theories have been proposed about the pathogenesis of CBC. First, the hydatid cyst progressively compresses the biliary tract wall, which causes necrosis and eventually CBC. Second, small biliary radicals in the pericystic wall generate high intracystic pressure, resulting in atrophy followed by the rupture of the biliary radicals.[10,12,13] Radiological findings suggesting the presence of CBC include deterioration of the integrity of the cyst wall and the presence of a

connection between the biliary tract and the hydatid cyst with cystic material in the biliary tract.[14] Ruptured materials in the biliary tract appear as various anechoic or hypoechoic shapes without acoustic shadowing on ultrasongraphy. On Computed tomography, the materials in the intrahepatic bile ducts can appear as high density linear structures. The loss of integrity of the cyst wall suggests CBC.[15]

Ultrasonography can detect a connection in 30%-74% of patients having major CBC preoperatively.[14] CT detects 75% of major CBC cases and allows for the evaluation of other organs.[16,17] ERCP and Magnetic Resonance Cholangiopancreaticography (MRCP) are ineffective in predicting pre-operative CBC because of the high pressure inside the cyst.[11] ERCP and MRCP can used pre-operatively for patients with obstructive jaundice, a history of cholangitis, or elevated liver enzymes. ERCP and MRCP demonstrate dilated bile ducts and daughter vesicles or a germinative membrane of hydatid cysts inside biliary radicals. If the bile ducts are evaluated with pre-operative ERCP, it is not necessary to do common bile duct exploration.[18] in our jaundice obstructive jaundice was present in 40% of the patients. Half of these patients underwent ERCP followed by elective laparoscopic surgery, while the other half underwent open surgery with CBD exploration. An elevated ALP as a predictor for CBC is controversial. After multivariate analysis, a history of jaundice, pre-operative ALT and ALP were found to be non-significant as predictors for CBC.[10,19]

Size of the cyst has always been reported as an important predictor for the presence of CBC. Atli et al, concluded that a cyst diameter >10.5cm was a significant predictor for frank rupture, while a cyst diameter of >14.5cm was signifcant for occult CBC.[6] Demircan et al, reported that a cyst diameter >8.5cm was a predictor for CBC.[9] Kilic et al, reported that a cyst diameter of >7.5cm was a risk factor for intra-operative bile leakage and postoperative biliary fistula.[10] In another study including 183 cases with occult and asymptomatic cysts, Unalp el al reported that a cyst diameter of more than 10cm predicted post-operative bile leak.[7] In our study, the cyst size of cases with CBC ranged between 6.4-10.8cms with a mean cyst size of 8cms. Other risk factors associated with an occult cystobiliary communication are male>female (40% vs. 10%), multiple cysts, multiloculated and degenerated cyst than unilocular, cysts near biliary bifurcation, presence of bile stained or purulent cyst contents.

Perdomo et al, reported that CBC is more common in cysts located close to the hilum, and that these hilar cysts are responsible for many serious complications.[20] In relating CBC to the Gharbi morphological classification of cysts, El Malki et al detected CBC frequencies of 27% and 31.7% in stage 3 and 4 cysts (multilocular and degenerated).[21] In our study, 70% of cysts are found in segment 4 and 5. Gharbi's type 4 cysts are seen in 12 patients while 8 patients had either type 2 or 3 cysts.

Pre-operatively diagnosed frank fistulae presenting with obstructive jaundice and/or cholangitis can be effectively managed by ERCP, which is both diagnostic and therapeutic and can be a scheduled elective surgery or open CBD exploration followed by partial pericystectomy with omentoplasty.[22,23] Some authors recommend routine ERCP in cysts larger than 7.5cm in diameter to assess the presence of CBF; however, there is no consensus supporting such a recommendation.[6,24]

There are several methods for intraoperative detection of CBC like leaving gauze in the cavity after opening the cyst, compressing gall bladder, intra-operative cholangiography, explore the cyst cavity with laparoscopic camera. Intraoperatively diagnosed CBF can be managed by several techniques. These include simple suturing or suturing with biliary decompression by the means of a transcystic tube (through the cystic duct after cholecystectomy) or a T-tube through a choledechotomy.[25,26] For major communication (>5mm) intraoperative cholangiogram is performed and if any filling defects are noted in CBD, exploration followed by T tube drainage is done.[27,28]

For minor communication (<5mm) direct suturing if cyst wall is not calcified, suturing with omentoplasty if the cyst wall is calcified.[27,28] Occult CBF not detected pre- or intra-operatively usually presents with post-operative biliary fistula. It is defined as persistent biliary drainage for more than 10 days post operatively (regardless of amount).[7] The reported incidence is 1-10% (1% in our series).[7] It can be low output (<300ml/day) or a high output (>300ml/day) fistula. The low output fistulas can be managed conservatively with external drainage, they usually resolve by 6 to 17 days. Endoscopic sphincterotomy is indicated in high output fistulas and in patients with prolonged biliary drainage (>3weeks).[9,29] ERCP with sphincterotomy ± stenting can be done with a success rate of 83 to 100% with resolution of biliary fistula in 2 to 4weeks time.[9,29]

CONCLUSION

Occult cystobiliary communication may be predicted preoperatively by using the predictors of ALP, total bilirubin, total leucocyte count, cyst size, cyst location and type.

In our experience conservative surgeries were found to be adequate in managing cystobiliary communication with minimal morbidity. Patients who develop postoperative biliary fistula with an output less than 300mL/day may be managed expectantly. Biliary fistula of more than 3weeks duration or with output exceeding 300mL/day requires endoscopic intervention.

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