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Original Research Article

A clinical study for preoperative prediction of cyst-biliary communications in hepatic hydatid cyst disease

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ABSTRACT

Background: Cyst-biliary communication with hepatic hydatid cyst disease is responsible for postoperative bile leakage after surgical management. This study aims to detect various predictors of cyst-biliary communication and their predictive accuracy.
Methods: This study was done in the patients of hydatid cysts who underwent surgical management for hydatid disease of the liver. Various factors were studied and their accuracy for preoperative prediction of cyst-biliary communication analyzed.
Results: There were 38 (22 males, 16 females) patients with hepatic hydatid cysts with a mean age of 38.7±15.4 years. Cyst-biliary communications were detected in 12 patients (31.6%). Independent strong predictors were tenderness in right hypochondrium (p=0.035), total leucocyte count (TLC)>12,000/mm^3 (p=0.0017), eosinophil count >5 × 108/l (p=0.0086), red blood cell distribution width (RDW) >15% (p=0.014), segment IV,V,VII involvement and cyst size >10 cms (p=0.01) on multivariate analysis.
Conclusions: Cyst-biliary communication is more common in patients presenting with tenderness in right hypochondrium, large cyst size, location in the central segments of liver close to biliary confluence, and with high values of TLC, Eosinophil count and RDW. The predictors demonstrated in this study should allow the likelihood of cyst-biliary communication to be determined preoperatively and, thus, indicate the need for additional procedures during operations to prevent the complications of biliary leakage.

Keywords: Cyst-biliary communications, Hepatic hydatid cyst, Hydatid cyst disease, Postoperative bile leak

INTRODUCTION

Hepatic infection with *Echinococcus granulosus* is a major public health problem worldwide and a common cause of cystic liver lesions in endemic areas. Infestation of humans with larval form incidentally causes hydatid cyst formation in different organs; most commonly in liver.[1] Hepatic hydatidosis is a significant health problem in Mediterranean and tropical countries such as Turkey, the Middle East, South America and Australia. In India, highest prevalence is reported from Andhra Pradesh and Tamil Nadu.[2] As an endemic disease; it causes social and economic losses.

Cyst-biliary communication (CBC) is reported in 5%-42% of hepatic hydatid disease cases. It is classified into two 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 postoperatively by an external biliary fistula.

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.[3]

Diagnosis of occult cyst-biliary communications is challenging because the symptoms and radiological findings in preoperative period are not clear. If the cyst-biliary communication is not be detected and repaired during surgery, postoperative biliary leakage and other biliary complications may occur in such cases often leading to increase in hospital stay and postoperative morbidity. Recognition of preoperative investigations and factors which can predict cyst-biliary communication therefore is very important in this setting.

METHODS

Study design

This study was conducted on surgically managed hepatic hydatid cyst patients in surgery department of RNT Medical College, Udaipur, Rajasthan from April 2012 to May 2019.

A specially designed detailed proforma was used to document and organize individual patient data which included clinical features, laboratory investigations, radiological and preoperative findings. The proforma was updated, during patient's hospital stay, during follow up and available hospital records.

Tenderness, total leukocyte count (TLC), eosinophil count, red blood cell distribution width (RDW), serum alkaline phosphatase, cyst size, cyst number, involved hepatic segment were analyzed for their predictive accuracy of cyst-biliary communications.

Statistical analysis

Descriptive and inferential statistical analysis was carried out. Results on categorical measurements were presented in number (%) and results on continuous measurements were presented on mean±SD (Min-Max). Chi-square/Fisher Exact test was used to find the significance of study parameters on categorical scale between two or more groups. A p value less than 0.05 was considered as significant.

RESULTS

Out of 38 cases of surgically managed hepatic hydatid cyst 12 (31.6%) had cyst-biliary communications. Mean age of presentation was 38.7±15.4 years. Disease prevalence was more common in males (22) as compared to females (16).

Pain in right hypochondrium was the most common presenting symptom (92%) (Table 1) Fever as a presenting symptom was observed in 7 patients, out of which 4 patients had cyst-Biliary communication (57%). Mass was a less common (6) presentation, but cyst-biliary communication was observed in 3(50%) of these patients. Tenderness in right hypochondrium was the most common sign observed in patient with hepatic hydatid cyst disease (76.3%). All patients with cyst-biliary communications, presented with tenderness in right hypochondrium, however only some (41.37%) patients with such tenderness had cyst-biliary communications (Table 1). Hepatomegaly was noted in 7 (18.4%) patients.

Table 1: Relation of cyst-biliary communication with tenderness right hypochondrium.

Factor	Value	No. of cases	Cyst-biliary communication detected	Cyst-biliary communication not detected	P value
Tenderness in right hypochondrium	Present	29	12	17	0.035 (S)
	Absent	9	0	9	

'NS' indicates not significant p value; 'S' indicates significant p value.

Table 2: Relation of cyst-biliary communication with blood indices.

Factor	Value	No. of cases	Cyst-biliary communication detected	Cyst-biliary communication not detected	P value
TLC (per mm^3)	>12000	9	7	2	0.0017 (S)
	<12000	29	5	24	
Eosinophil count per litre	>5×10^8	13	8	5	0.0086 (S)
	<5×10^8	25	4	21	
RDW	>15%	14	8	6	0.014 (S)
	11-15%	24	4	20	
Serum alkaline phosphatase (IU/l)	>140	11	4	7	0.71 (NS)
	20-140	27	8	19	

'NS' indicates not significant p value; 'S' indicates significant p value.

Majority of patients presented with normal total leucocyte count (76.31%). Out of 9 patients who had leukocytosis (TLC>12000/mm^3), cyst-biliary communications were detected in 7 patients. Majority of patients presented with normal eosinophil count (65.8%). Cyst-

biliary communications were noted more commonly when eosinophil count was above normal limit. RDW was normal in 24 (63.1%) patients. Out of these 24 patients, only in 4 patients (16.7%) cyst-biliary communication was detected, whereas among 14 patients with raised RDW 8 patients had cyst-biliary communication (57.14%) (Table 2).

Twenty-seven patients out of 38 presented with normal serum alkaline phosphatase, 8 (29.6%) of which had cyst-biliary communication. Only 4 (36.36%) of 11 patients with raised serum alkaline phosphatase had cyst-biliary communication (Table 2).

Twenty-five (65.79%) patients presented with hepatic hydatid cyst of size ranging from 5-10 cms. Only 1 (2.6%) patient presented with cyst size less than 5cms. However, maximum number of cyst-biliary communications were seen in cysts larger than 10 cms. Majority of patients (29) presented with single hepatic hydatid cyst and only in 7 (24.13%) of these cyst-biliary communications were detected intraoperatively. Nine patients (23.7%) presented with multiple hepatic hydatid cyst and 5(55.5%) of them had cyst-biliary communications. Most of the cysts (68.4%) were seen in right lobe of liver. More than one segment was involved in 24 (63.15%) patients. Maximum number of cyst-biliary communications were found in cysts located in segments IV (100%), V (75%), IV/V/VIII (80%) implying thereby that these communications are more likely to be detected in cysts present in segments near the biliary confluence (Table 3).

Table 3: Relation of cyst-biliary communication with size, number and location of cyst.

Factor	Value	No. of cases	Cyst-biliary communication detected	Cyst-biliary communication not detected	P value
Cyst size in cms	>10	12	8	4	0.01 (S)
	5-10	25	4	21	
	<5	1	0	1	
Number of cysts	Single cyst	29	7	22	0.10 (NS)
	Multiple cyst	9	5	4	
Involved segment of liver	Segment IV	1	1	0	
	Segment V	4	3	1	
	Segment VI	3	0	3	
	Segment VII	5	0	5	
	Segment VIII	3	0	3	
	Segment IV/V/VIII	5	4	1	
	Segment VI/VII	5	2	3	
	Segment IV/VI/VII	3	0	3	
	Segment V/VI/VII	5	2	3	
	Segment VII/VIII	4	0	4	

'NS' indicates not significant p value; 'S' indicates significant p value.

DISCUSSION

Echinococcosis is a severe parasitic disease which affects both animals and humans, with multiform complications and frequent relapse. 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 into intra-biliary tree. Since cyst-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. Cyst-biliary communication may be due to either the compression effect of cyst-over biliary tract, which causes necrosis and eventually cyst-biliary communication or small biliary radicals in the peri-cystic wall.

Preoperative prediction of cyst-biliary communication has crucial role in management of associated morbidity, especially occult ones. Studies have suggested cyst size, number of cysts, localization of cyst, calcified wall, recurrent disease, preoperative jaundice, and preoperative elevated serum alkaline phosphatase (ALP) as predictor for cyst-biliary communications.[4]

In this study group, tenderness in right hypochondrium was noted to be statistically significant predictor of cyst-biliary communication in hepatic hydatid cyst disease (p=0.035). Tenderness may be due to secondary infection of the cyst resulting in local inflammation, which is more likely when cyst-biliary communications were present.

Recent studies investigating predictive variables for cyst-biliary communications have generally evaluated TLC as an inflammatory marker. Majority of patients of hepatic hydatid cyst have been reported to present with normal TLC, which is similar to this study also where 29 (76.21%) patients presented with normal TLC. TLC has

been defined as independent risk factor in some studies whereas other studies have not found any relationship between TLC and cyst-biliary communication.[5-9] In this study group, it was observed that TLC was statistically a strong predictor of cyst-biliary communications (p=0.0017). TLC is likely to be elevated in case of secondary infection of cyst, which is more likely when cyst-biliary communications are present.

Absorption of hydatid antigen following cyst rupture into biliary channels or the peritoneum may increase the eosinophil count. Eosinophilia has been reported in 29%-100% of patients with intra-biliary rupture.[10-13] In this study group, majority of patients presented with normal eosinophil count (65.8%). However, raised eosinophil count (>5×108/l or >6%) wasseen more commonly in patients with cyst-biliary communications, making it an independent risk factor for cyst-biliary communication in this study. Humayun and colleagues11 found eosinophilia (>10%) more commonly in the patients with cyst-biliary communication. Eosinophilia (>3%) may be found in 25% to 45% of patients with hydatid cyst in Western countries, but this is a non-specific finding in endemic areas.

Additionally, Meneske found that RDW was significantly higher in patients with cyst-biliary communication than in patients without cyst-biliary communications.[14] Chronic inflammation due to occult cyst-biliary communication could be the source of this relationship. In this study, majority of patients presented with normal RDW (64%). Cyst-biliary communication was observed more in patients who presented with higher RDW (57.14%) as compared to patients with normal. Higher value of RDW was statistically significant predictor of cyst-biliary communications (p=0.014). A positive correlation has been found between RDW and inflammatory markers such as C-reactive protein and the erythrocyte sedimentation rate (ESR) in various studies.[15]

Even a large hydatid cyst of liver may not alter liver function tests, and transaminase levels are usually normal. Cholestatic enzymes, such as alkaline phosphatase can be mildly elevated in about one third of patients, especially in patients with biliary compression. Preoperative alkaline phosphatase level has been reported to be a strong predictor of cyst-biliary communications.[16] However Meneske found that serum alkaline phosphatase was not a statistically significant predictor of cyst-biliary communication (p>0.05).[14] Similarly in this study, alkaline phosphatase was not found statistically significant predictor of cyst-biliary communication (p=0.71). These different results may be dependent on several reasons such as the inclusion of different patient groups in different studies, nonspecific hepatobiliary enzyme studies for hepatic hydatid cyst disease and cyst-biliary communication, especially ALP levels, which is not originating exclusively from the liver.

Size of the cyst has always been reported as an important predictor for the presence of cyst-biliary communication in various studies.[7,8] The hypothesis is that the increase in the cyst size is associated with an increase in the intra-cystic pressure causing pressure necrosis of the adjacent biliary radicles with a subsequent rupture of the cyst into the biliary system. Similarly, in this study group, cyst size was found to be a statistically strong predictor of cyst-biliary communication (p=0.01).

Multiple cysts on presentation are considered as a risk factor for cyst-biliary communication in hepatic hydatid cyst disease. Nakeeb concluded in his study that presence of multiple cysts was statistically significant risk factor of cyst-biliary communication (p value=0.006).[16] However multiple cystic presentation was not found statistically significant predictor of having cyst-biliary communication (p value=0.10) in this study group. Similarly, Demarcan concluded that the frequency of biliary leakage was not affected by cysts being single or multiple (p=0.84).[8]

The segment involved has been considered important in predicting cyst-biliary communications. Perdomo et al reported that cyst-biliary communication was more common in cysts located close to the hilum, and that these hilar cysts are responsible for many serious complications.[17] Likewise, Kayaalp et al found that cysts located close to the hilum tended to rupture into the biliary system.[2] In a study by Alan, although cyst-biliary communications were more common in cysts located close to the hilum than cysts far from the hilum (36.9% vs 35.3%, respectively), this difference was not statistically significant.[18] Similarly in this study group, 24 patients out of 38(63.15%) involved more than one segment. Maximum number of Cyst-biliary communication were found in cysts in segmentV (3 out of 4 as a single cyst (75%)) (in 9 patients out of 14 as a part of multiple cystic lesion (64.2%)), IV/V/VIII (in 4 patients out of 5(80%)), segment IV (1 out of 1). No cyst-biliary communication was detected when isolated hydatid cyst involved the segment VI, VII, VIII (none of the 11 patients). The more common presence of cyst-biliary communication in cysts located close to the biliary confluence or hilum may be related to the number and density of perihilar biliary tracts in these areas.

The management strategy of cyst-biliary communication depends on when it is diagnosed. 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. Some authors recommend routine ERCP in cysts larger than 7.5 cm in diameter to assess the presence of cyst-biliary communication; however, there is no consensus supporting such a recommendation.[19] Intraoperatively diagnosed cyst-biliary communications can be managed by several techniques. These include simple suturing or suturing with biliary decompression by the means of a

trans-cystic tube (through the cystic duct after cholecystectomy) or a T-tube through a choledochotomy.

In this study, cystectomy with partial pericystectomy with external tube drainage was the commonest procedure done and primarily closure of all intraoperatively detected cyst-biliary communications could be done, as in this study all cyst-biliary communications were small. Among the 38 patients who were managed surgically for hepatic hydatid cyst, only 3(7.9%) has been developed postoperative biliary complications. All 3 patients were managed conservatively with external tube drainage.

CONCLUSION

Preoperative prediction of cyst-biliary communication has crucial role in management of associated morbidity, especially in occult cyst-biliary communication. In this study, authors studied tenderness, TLC, eosinophil count, RDW, alkaline phosphatase, cyst size, cyst number, involved hepatic segment for predictive accuracy of cyst-biliary communications. Tenderness in right hypochondrium ($p<0.05$), TLC>12000/mm^3 ($p<0.05$), eosinophil count >5×108/l ($p<0.05$), RDW>15% ($p<0.05$), cyst size ($p<0.05$) and involvement of segment near biliary confluence were found statistically significant predictors of cyst-biliary communications in hepatic hydatid cyst disease.

The predictors demonstrated in this study should allow the likelihood of cyst-biliary communication to be determined preoperatively and, thus, indicate the need for additional procedures during operations to prevent the complications of biliary leakage.

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