



**ORIGINAL RESEARCH PAPER**

**Pediatric surgery**

**PREDICTORS OF CYSTOBILIARY COMMUNICATION IN HYDATID DISEASE OF LIVER, INTRA-OPERATIVE AND POST-OPERATIVE MANAGEMENT IN CHILDREN AT A SINGLE INSTITUTION IN NORTHERN INDIA.**

**KEY WORDS:** Cystobiliary communication (CBC), Intra-biliary rupture.

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**ABSTRACT**

**Introduction:** Hydatid disease in children is highly prevalent in this part of world due to lack of proper hygiene. Cystobiliary communication(CBC) with hepatic hydatid disease is responsible for postoperative bile leakage and morbidity.

**Aims:** Our study aimed to predict the communication, intraoperatively diagnose it and manage it postoperatively in case not diagnosed previously.

**Material and Methods:**This is a prospective , cohort study of all children with hydatid disease of the liver. Patient data were recorded on an internal web-based registry system supplemented by paper records. All the patients who underwent surgery for hydatid disease from 2013 to 2019 were assessed for presence of CBC and only patients with CBC were included in the study and studied.

**Results:** A total of 97 patients presented with hydatid disease of liver during the said period. There were a total of 20 (20.6%) CBC among these .Males had more CBC than females (m:f=2:1).

**Conclusion:** Cyst size >10cms, jaundice, right lobe cyst preoperatively were the most important predictors of CBC.It was found that on-table suturing of CBC with deposition of pedicled omentum was the most effective way of preventing post-operative bile-leak. Management depends on the size of the fistula, the site of the cyst, and the experience of the hepatobiliary surgeon.

**INTRODUCTION:**

In endemic countries, up to 25% of hydatid disease is reported in children [1,2]. The symptoms of hydatid disease are protean and depend on which organs are affected. The most commonly affected organ is the liver in adults, though lung and liver are nearly equally involved in children[3,4]. Hepatic hydatid disease is usually presented by right upper abdominal pain, hepatomegaly, or a palpable mass. As the cyst enlarges, the risk of cyst wall dehiscence with a subsequent rupture increases. The cyst may rupture into the peritoneal cavity, pleural cavity, blood stream or, most commonly, the biliary system, which causes cyst biliary fistula (CBC) [5]. Cystobiliary fistula is reported in 5%-42% of hepatic hydatid disease cases [6,7,8].

Intra-biliary rupture could be an overt frank or silent occult [9]. In overt situations, a major cyst contents, like debris, daughter cysts and fragments of the laminated membrane may go down into the common bile duct [10]. Most CBCs are occult in nature without specific clinical, radiological or laboratory findings due to relatively small communication and discovered only during or after surgery as a postoperative biliary fistula (PBF) [11,12]. Bile usually not seen in the cyst cavity in cases of occult CBC due to high intracystic biliary pressure gradient (80 vs. 20 cm H<sub>2</sub>O subsequently). After cyst evacuation, bile flows back into the cyst due to reversed pressure gradient [13,14].

In this instance, undesired complications could be happen such as prolonged biliary leakage, biliary peritonitis and abscess, an increased morbidity, cost and hospital stay will ensure. Our study was aimed at predicting the possibility of such CBC's, possible intraoperative diagnosis and treatment and post-operative management.

**Patient and Methods:**

This was a prospective comparative observational study

conducted in the Department of Pediatric surgery at single institution in Northern India. The study was conducted over a period of 6 years from June 2013 to June 2019 with new cases included till June 2017, thus having a minimum follow-up of more than 2 years. All patients were treated with albendazole 15mg/kg/d for at least 4 weeks preoperatively and continued postoperatively for minimum 3 cycles with each cycle extending up to 3 weeks with 1-week gap in between which was in accordance with the study by Morris et al [15]. Follow-up was conducted for a minimum of 2 years and ultrasound abdomen was performed at 6 monthly intervals to look for any recurrence.

If laproscopic enucleation of hydatid after initial laparoscopic evaluation, the suitability of the cyst for laparoscopic management was confirmed. Essentially the following steps were adopted: peri cystic packing with cetrimide soaked or betadine soaked gauze to take care of spillage, decompression of the cyst by aspiration using a wide bore needle through one of the 5-mm ports with placement of two 5-mm suction cannulas next to the aspirating needle to control the spillage, naked eye examination of the fluid for the presence of bile or pus, injection of 3% hypertonic saline or cetrimide for 10 minutes to ensure complete killing of the organism, followed by aspiration. Cystotomy was made in the pericyst in nondependent area with scissors or with the hook electrode, followed by removing the germinative membrane in a plastic bag or by using locally improvised specimen bags to prevent contamination followed by extraction through the epigastric port. Cyst cavity was telescoped for any remaining membranes or biliary communications, in case of overt signs of cyst-biliary communication, use of a scolicalid agent was avoided and after marsupialization, the opening was sutured with 3-0 Vicryl suture. The management of the residual cavity was achieved by placement of omentum into the residual cavity if the location or the configuration of the cyst warranted

or by simple drainage, unroofing, peri cystectomy or other techniques. Sometimes another cystotomy was made in the cyst in a dependent area for drainage of the cavity. During the procedure, spillage of cyst contents was anticipated and rated for by an independent observer.

In open technique a right subcostal approach was used in most patients. Same precautions were taken to avoid spillage as in laparoscopic method. At the end of the procedure, the cavity was examined for any bile duct leakage which, if found, was closed with vicryl suture. The residual cavity was finally managed by mainly either of the two techniques: External tube drainage or Omentopexy.

**RESULTS:**

Over the study duration (2012-2016), 97 cases of paediatric hydatid disease liver were operated in our department. These patients were operated and total of 20(20.6%) patients turned out to have CBC while as 77 (85.5%) patients finally had no CBC. These 20 patients were studied to look for any preoperative predictors and their management and were compared with those without a fistula. The data is tabulated and analysed below.

Variables	Total	With biliary communication 20 (20.6%)	Without biliary communication 77 (85.5%)
Male	69(72%)	12(60%)	50(64.9%)
Female	25(28%)	8(40%)	20(25%)
Pain	90(92%)	20(100%)	70(90%)
Preoperative obstructive jaundice	5(5%)	3(15%)	2(2.5%)
Preoperative raised liver enzymes	10(10.3%)	7(35%)	3(23%)
Cholangitis	1(1.03%)	1(5%)	0(0%)
Size of cyst <10cms	80(82.4%)	4(20%)	76(98.7%)
Size of cyst >10cms	17(17.8%)	16(80%)	1(1.1%)
Right lobe cyst	80(82.4%)	15(75%)	65(84.4%)
Left lobe cyst	17(17.5%)	5(25%)	12(15.5%)
Management of cavity by omentoplasty	48(49.4%)	15(75%)	33(42%)
By tube drainage	49(50.5%)	5(25%)	44(57%)
Postoperative ERCP	1(1.03%)	1(5%)	0(00%)
N=97			

**DISCUSSION:**

CBC was found in 20 of 97 patients of hepatic hydatid operated in our centre, i.e 20.6%. It has been reported to be present in up to 40% of the cases of hepatic hydatid disease and is responsible for most of the post-operative complications [6-8, 15, 16].

Since this communication can result in a postoperative bile leak, persistent biliary drainage, biliary peritonitis so it 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. Preoperative predictors are most vital to help manage a CBC. These predictors are got from a thorough clinical examination, liver function tests, radiology, and intraoperative findings.

In our series, obstructive jaundice was present in 15% of the patients with CBC, which was similar to what Atli et al. [7] who reported obstructive jaundice in 25% patients in his study.

Although the pre-operative levels of ALT and ALP were significantly higher in the CBC group, however, after

multivariate analysis, a history of jaundice, pre-operative ALT and ALP were found to be non-significant as predictors for CBC, which is consistent with recent reports [14, 17].

The size of the cyst has always been reported as an important predictor for the presence of CBC. The hypothesis is that the increase in the cyst size is associated with an increase in the intracystic pressure causing pressure necrosis of the adjacent biliary radicles with a subsequent rupture of the cyst into the biliary system [18]. However, the cut off value for the cyst diameter that predicts CBC has not been settled. In 2001, Atli et al. [7] analyzed the data of 116 patients, and 24 had CBC.

They concluded that a cyst diameter >10.5 cm was a significant predictor for frank rupture, while a cyst diameter of >14.5 cm was significant for occult CBC. Kilic et al. [17] reported that a cyst diameter of >7.5 cm was a risk factor for intra-operative bile leakage and postoperative biliary fistula. In another study including 183 cases with occult and asymptomatic cysts, Unalp et al. [19] reported that a cyst diameter of more than 10 cm predicted post-operative bile leak. In the current study, we also found that the cyst diameter of >10cms was a significant predictor for the presence of CBC.

One patient in our study had a severe postoperative cholangitis. This patient had an occult CBC and required an ERCP in postoperative period. Right lobe cysts have a greater predilection for CBC.

**CONCLUSION:**

Cyst size >10cms, jaundice, right lobe cyst preoperatively were the most important predictors of CBC. It was found that on-table suturing of CBC with deposition of pedicled omentum was the most effective way of preventing post-operative bile-leak. Management of communication depends on the size of the fistula, the site of the cyst, and the experience of the hepatobiliary surgeon.

**Declarations:**

The study was approved by institutional ethical committee of Sheri Kashmir Institute of Medical Sciences, Soura, Srinagar, India.

A proper informed consent was taken from the parents and/or legal guardians of all the patients as they were minors.

The procedures were carried out in accordance with relevant guidelines and regulations.

The study was an observational study on hospital patients and a proper consent was taken before any procedure was being contemplated as per ethics and rules framed by the ethical committee.

The study was not funded as the procedure is well established in the department and it was an observational study.

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