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Original Article

Outcome of Surgically Treated 79 Patients of Hepatic Hydatidosis: A Single Center Tertiary Care Hospital Experience in Bangladesh

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Abstract

Background: Proper patient selection for most appropriate treatment option for hepatic hydatid is crucial to ensure successful treatment outcome. Chemotherapy, newer approaches, like PAIR procedure, (puncture, aspiration, injection and reaspiration) laparoscopy are available. As the indications for these approaches are restricted, surgery remains the key treatment strategy with hope for complete cure. Radical surgical excision coupled with chemotherapy and long-term aggressive chemotherapy for partially resected ensures lowest morbidity and mortality.

Methods: This cross sectional observational study of 15-year single-institution experience, intended to further validate different surgical procedures in management of hydatid disease of liver and its outcome. Patients were randomly selected irrespective of their age, sex and mode of presentation. Different preferred surgical treatment modalities, perioperative complications, recurrences, and length of hospital stay were retrospectively analyzed.

Result: The study was carried out on 79 Bangladeshi patients diagnosed hepatic hydatidosis, treated surgically from January 2002 to December 2017 in BIRDEM General Hospital. Among them was no anastomotic leakage or mortality in the immediate postoperative period in our series.

Conclusion: This study revealed, proper case selection and consideration of risks-benefits, indications-contraindications for each case of Hydatid cyst of liver, for making a decision for type and timing of surgery is key to successful outcome. Surgery is the mainstay of treatment strategy. Radical surgery is key to effective and successful outcome, ensures better quality of life and prevents recurrence of the disease.

Key word: Hydatid cyst, Pericystectomy, partial cystectomy, hepatic hydatid, SOL Liver

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Introduction:

Human echinococcosis is a zoonotic infection caused by tapeworm of *Echinococcus* genus. Appropriate patient selection for most applicable treatment option for hepatic hydatidosis, ensures lowest recurrence with minimal morbidity and mortality. Surgery is the mainstay of treatment for Hydatid cyst of liver. 15-year single-institution experience, intended to further validate different surgical procedures in management of hepatic hydatid disease. Radical surgery is key to effective and successful outcome, ensures better quality of life and prevents recurrence of the disease.

Methods:

This is a retrospective cross sectional observational study of 79 patients of hepatic hydatidosis, treated surgically from January 2002 to December 2017

in BIRDEM General Hospital, a major specialized tertiary care hospital in Bangladesh. Patients were randomly selected irrespective of their age, sex and mode of presentation. Upper abdominal fullness or heaviness with discomfort, occasional pain, abdominal mass and occasional fever were mode of presentations. Ultrasonography of abdomen was the primary diagnostic tool supplemented with Enzyme-linked immunosorbent assay (ELISA) and Indirect haemaglutination assay (IHA). CT scan was for mapping the disease and planning surgery. Apart from clinical parameters, we analyzed surgical treatment modalities, perioperative complications, recurrences, and length of hospital stay.

Surgical Approaches:

Radical surgery for complete removal of the cyst or diseased segment was the cornerstone of surgical management. It includes, Pericystectomy (figure 1, 2 & 3), anatomical (figure 4) and nonanatomical hepatic resections. When the cyst is large, deep-seated, occupies more than two hepatic lobes, overlies hepatic hilum or difficult in location or position; partial cystectomy with safe evacuation of cyst contents, preventing spillage, application of scolicidal agent and management of residual cavity followed by marsupialization and omentoplasty was done. Partial cystectomy aimed at organ preservation and prevention of adjacent tissue damage. Tissues were histopathologically studied. CBD exploration and biliary reconstruction was done in selected cases. All cases received albendazole perioperatively according to protocol.

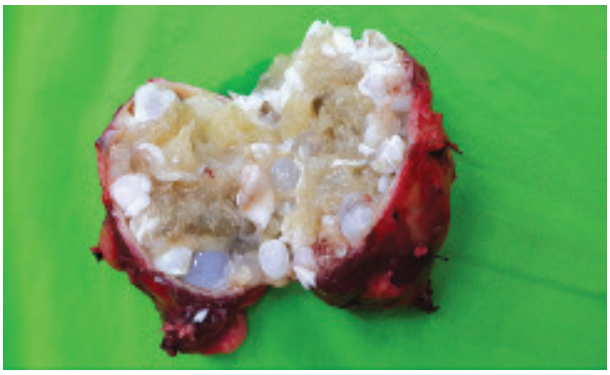

Figure 1. *Pericystectomy specimen with daughter cysts (by closed technique)*

Figure 2. *Pericystectomy specimen after removal of daughter cysts (by open technique)*

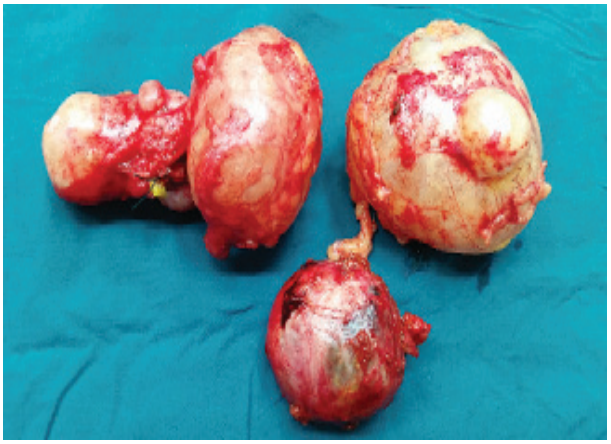

Figure 3. *Intraperitoneal cysts in dessiminated hydatid cyst*

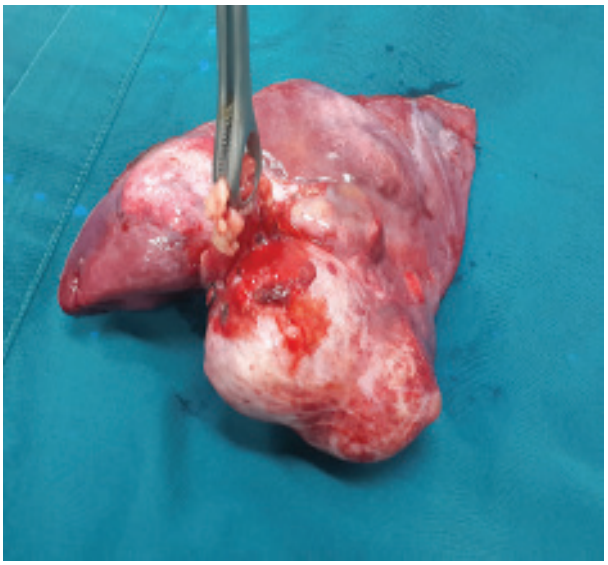

Figure 4. *Left Hepatectomy specimen with multiple hydatid cyst*

Post-operative period:

Patients were thoroughly evaluated considering objective and subjective complaints. Started oral feeding between 2nd to 5th postoperative days and discharged between 7th to 21st postoperative days. Diabetic patients had more frequent follow-ups and some needed special care. Good glycemic control was maintained. They were followed up till December 2017 with at least four times in outpatient.

All patients received postoperative chemotherapy, single cycle of 4-6 weeks for 3 – 6 cycles with two weeks interval. They were followed up with abdominal USG three monthly for first year and then yearly.

Post-operative complication:

There were minor and major as well as intra and postoperative complications. There were hemorrhage, intra-abdominal collection, wound infections, minor biliary leak, recurrence after 36 months of follow up and were managed according to evaluation verdicts. There is no immediate postoperative mortality in our series.

Result:

This single center, cross sectional observational retrospective study, conducted on 79 patients revealed female dominance with 17 male and 62 female with ratio of M: F = 1.0: 3.65. Patients ranging from 06 – 83 years with mean age at presentation was 34.6 years (Table 1).

Table 1. *Demographic data*
N = 79 Male 17 Female 62 M : F :: 1.0 : 3.65

Sex Age (yrs)	Male (%)	Female (%)
0 – 10	0	0
11 – 20	0	3
21 – 30	3	23
31 – 40	5	17
41 – 50	2	9
51 - 60	3	5
61 – 70	4	4
71 – 80	0	1

56 (70.89%) patients presented with heaviness & discomfort with occasional pain in upper abdomen, abdominal fullness & palpable upper abdominal mass, anorexia and history of non-specific low-grade fever (Table 2), which were present in 74% and 55% of the cases, respectively. 14 patients were complicated at presentation with recurrent disease in 07 cases, four with abscess in the cyst with sepsis, two with metastatic cysts in peritoneal cavity and one patient presented with obstructive jaundice – ERCP for biliary decompression revealed debri and daughter cysts in the CBD.

Table 2. *Presentation*

Presentation	n	%
Asymptomatic	9	11.39
Symptomatic	56	70.89
• Pain & heaviness in upper abdomen	56	
• Abdominal fullness & palpable abdominal mass	38	
• Low grade fever of varying duration	27	
• Anorexia	27	
• Occasional Vomiting	16	
Complicated	14	17.72
• Infected	06	
• Ruptured (in the biliary tree)	01	
• Recurrent	07	

Majority of patients in our series presented with right lobe lesion with 52 (65.82%) patients in the right lobe, 14 (17.72%) in left lobe, 13 (16.46%) had bilober involvement and among them two had peritoneal extensions (Table 3). Seven patients at presentation had recurrent cysts. All cases were treated with albendazole (400 mg twice daily for adults & 100 – 200 mg for children) for two weeks before surgery to reduce infectivity and risk of recurrence of the cyst.

Table 3. *Location of cysts (USG and CT scan)*

Location	Lesion	n	%
Right lobe	Single	43	54.43
	Multiple	09	11.39
Left lobe	Single	14	17.72
	Multiple	-	-
Both lobes	-	11	13.92
	Metastatic cysts in peritoneal cavity	02	2.53

In 36 patients partial cystectomy (fig. 1) and omentoplasty (fig. 2) were done; among them eight patients had biliary communications with the cyst which were closed by prolene sutures. 12 cases of right lobe cysts were managed by pericystectomy (fig. 3), 05 by non-anatomical hepatic resection and 03 by hepatic segmentectomy (segments VI & VII) (fig. 4). Hepatic lobectomy (fig. 5) was performed in 23 cases of left lobe cysts. In all cases drain/s were placed accordingly. Samples were sent for histopathological examination.

Table 4. *Operative procedures*

Operation	n	%
Partial Cystectomy & omentoplasty	36	45.57
Pericystectomy	12	15.19
Non-anatomical hepatectomy	05	6.33
Hepatic Segmentectomy (Right lobe)	03	3.80
Hepatic Lobectomy(Left lobe)	23	29.11

*peritoneal hydatid cysts were enucleated

Early postoperative period was uneventful. 13 (16.46%) patients had minor to major surgical wound infection with partial wound dehiscence in two cases and was dealt with accordingly. 06 (7.59%) patients had minor bile leak - <100 ml in 24 hours. Patient recovered with conservative measures. In all cases albendazole was continued postoperatively according to protocol. Routine histopathology of sent sample revealed no malignancy and features were compatible with hydatid cyst wall or cysts. There was no immediate postoperative mortality in our series.

All the patients were followed up according to protocol. 56 (70.89%) patients followed up for a maximum period of 60 months and with minimum four followups. During the period we had four cases of recurrence of hydatidosis after 36 months and they were managed according to evaluation results.

Table 5. *Postoperative course*

Parameters	n	(%)
Wound infection	13	16.46
Bile leak	06	7.59
Recurrence	04	5.06
Mortality	Nil	0.0
Follow up (up to 60 months)	56	70.89

Discussion

Hippocrates recognized human hydatid over 2,000 years ago and described it as 'liver full of water'. Dogs and other canines are definitive hosts and humans are accidental intermediate hosts. It is caused by larva of dog tapeworm. *E. granulosus* results in development of one or multiple unilocular hydatid cysts that in humans develop mainly in the liver (70%), lungs (20%) and 10% of cysts may occur anywhere in the body[1, 2].

Hepatic hydatidosis is endemic in the Mediterranean region, South America, Australia, China, Middle East and Eastern Europe, the incidence has declined significantly[1,2]. However, it is re-emerging in some areas with alarming increase in Bulgaria and Kazakhstan, and China[2,17]. It is a non-endemic zoonosis in Bangladesh. There is no specific data available; yet it is not uncommon in hospitals. In our series, we have surgically treated 79 patients from January 2002 to December, 2017 in BIRDEM General Hospital with Male : Female ratio 1 : 3.65, mean age was 34.6 years, the disease is three times more common in female.

Most cases, there is only one cyst, whereas in some cases, multiple cysts may be present (20–40%). Symptoms depend on the size and the number of cysts and possible compression of surrounding structures[19]. The hydatid cysts are slow growing approximately 2-3 cm / year and remain asymptomatic and undiagnosed for long time. The fluid inside the cyst is highly antigenic may cause severe anaphylaxis and death[3-5]. 56 (70.89%) patients presented with heaviness & discomfort with occasional pain in upper abdomen, abdominal fullness & palpable upper abdominal mass, anorexia and history of non-specific low-grade fever (Table 2) that were present in 74% and 55% cases respectively. 14 patients were complicated at presentation with recurrence in 07 cases, 04 with abscess with sepsis, 02 with metastatic peritoneal cysts and 01 presented with obstructive jaundice who required ERCP for biliary decompression that revealed debri and daughter cysts in CBD.

Routine laboratory hematological and biochemical investigations showed nonspecific Leukocytosis may suggest infection of the cyst. Eosinophilia is present in 25% cases, while hypogammaglobinemia is present in 30%. Mild to moderate elevation of liver enzymes like bilirubin and/or alkaline phosphatase level was noted in biliary encasement. Serological investigation

– Indirect hemagglutination assay (IHA) and the enzyme-linked immunosorbent assay (ELISA) have a sensitivity of 80% overall (90% in hepatic hydatid, 40% in pulmonary hydatid)[3-6]. ELISA was 84.2% sensitive and most specific test for hepatic hydatidosis. Sensitivity to eosinophilia or serological tests is low due to dead parasite, reduced absorption of antigen and impaired immune reaction.

USG is the gold standard for diagnosing hepatic hydatids (least invasive, least expensive and more informative). The Gharbi and WHO classification of hepatic hydatid cysts in USG is useful [3]. Hydatid cyst containing daughter cysts give a typical honey-comb appearance or water-lily sign, early cases show clear cystic fluid with sandy precipitates. Findings vary from purely cystic to solid appearing pseudotumors. Typical findings are not always present. Hydatid cysts of liver can cause complications in about 40% of cases[4] . Mechanical pressure effects by the expanding cyst, rupture of the cyst into the body cavities and into the biliary system, infection, sepsis, calcification and dissemination of hydatidosis. Hydatid cysts are also complicated by multiplicity and suppuration like a pyogenic abscess[5,6,7]. USG sensitivity is 85% to 95%, CT sensitivity is 95%. Literature review suggests MRI is more informative than CT scan . But we found Multiaxial CT scan was appropriately informative and accurately delineates the liver lesions. Primarily infected hydatidosis is relatively common, with frequency between 5% and 40%[8,9,10,18]. Majority of patients in our series presented with right lobe lesion with 52 (65.82%) patients in the right lobe, 14 (17.72%) in left lobe, 13 (16.46%) had bilober involvement and among them two had peritoneal extensions (Table 3). In our series two female patients were in their mid-pregnancy when diagnosed. They were operated after pregnancy was over for persistent cystic lesions.

Early uncomplicated type I & II cysts resolve by medical treatment with benzimidazole derivative; Albendazole and Mebendazole; Although albendazole 10 – 15 mg/ Kg of body weight in two divided doses or 400 mg twice daily for three to six cycles of one month of medication with two weeks interval is more preferred [7,9]. Medical treatment is contraindicated in the following: (1) large cysts, (2) cysts with multiple septa divisions (honeycomb-like cysts), (3) cysts prone to rupture (superficial), (4) infected cysts, (5) inactive cysts, (6) calcified cysts, (7) severe chronic liver disease, (8) bone marrow depression, and (9) early pregnancy[6,7].

Hydatid cyst of liver is composed of three layers: Adventitia or pericyst, comprises of host tissue consisting of compressed liver parenchyma and fibrous tissue, easily separable acellular white elastic Laminated membrane or ectocyst layer, the highly active germinal epithelium or endocyst layer lining the cyst then the hydatid fluid and brood capsules[10,11].

Radical procedures have lower rate of complications and recurrences but are technically difficult and often the risk of pedicle injury. The radical surgical procedure includes: Pericystectomy, Lobectomy and Non-anatomical Hepatectomy[12,13,14]. We did partial cystectomy and omentoplasty for large (> 15.0 cm) and intraparenchymal hydatid cysts. Omentoplasty has been advocated for its absorptive capacity of residual fluid in the cystic cavity. It stimulates macrophage migration into the operated area[15,22]. However, risk of anaphylaxis and peritoneal dissemination may have been overestimated. The pericyst is protective, prevents extension of infective process to liver parenchyma[16,17,18]. Redical excision or resections are technically demanding, but ensure complete and safe removal of the cyst [19,20,21].

We operated seven patients of recurrent hepatic hydatidosis (six months to one year after previous surgery) – two of them had metastatic cysts in the peritoneal cavity. Intraperitoneal cysts were enucleated from the mesenteric folds (Figure 3). Complicated cases with sepsis and abscess formation were treated with combination antibiotics according to culture and sensitivity. Simultaneous presence of hydatid cysts in two organs occurs in 5–13% cases[22,23]. In endemic areas, cysts elsewhere within the peritoneal cavity should be considered. However thorough imaging examination and surgical excision plays the key role. Intrabiliary rupture and cyst super infection are considered to be the most common complications of hepatic hydatidosis[18,19,20]. This patient was treated with partial cystectomy, closure of biliary communication with prolene suture and Roux en Y hepatico-jejunostomy after thorough irrigation of biliary tree by 20% saline. Prevention of spillage of cyst contents during surgery and intermittent irrigation of surgical field with 20% saline or 1% povidone iodine are important in preventing postoprative recurrence of hydatidosis. Patients who have jaundice or a history of cholangitis, elevated liver enzymes, and dilatation or debris in major bile ducts should be assessed for main bile duct contamination[21]. In 36 patients partial

cystectomy, marsupialization and omentoplasty were done; eight of them had biliary communications which were secured. 12 cases of right lobe cysts were managed by pericystectomy (fig. 1, 2&3), 05 by non-anatomical hepatic resection and 03 by hepatic segmentectomy. Hepatic resection was performed in 23 cases of left lobe cysts (fig. 4). 13 patients had parietal wound infections among them two had partial wound dehiscence were dealt with accordingly. 06 (7.59%) patients had minor bile leak <100 ml in 24 hours – stopped spontaneously after 8th postoperative day.

Patients were followed up by Ultrasonography of whole abdomen three monthly for first year and yearly onward. 56 patients attended up to 60 months of follow up among them four patients had recurrence detected after 36 weeks of follow up – treated with reoperation under cover of albendazole. Over the ages despite implementation of different surgical techniques and methods in our series, postoperative morbidity and mortality have not changed and it is individualized. Postoperative complications ranges from 19-37%[18,24]. We have realized that treatment failures commonly resulted from catheter obstruction by purulent material or from untreated loculations within the abscess. In our series, repeat laparotomy was performed on 5 patients, approaching a rate of 6.5%. Decisive factor in low rates of recurrence is the cautious surgical technique. Sepsis, advanced age, and coexistent serious general conditions were implicated as the cause of death in two of our patients, leading to a mortality rate of 2.6%. Our study is subject to a number of limitations inherent in the use of retrospective administrative data.

Conclusion

Treatment of hepatic hydatid disease is individualized. Appropriate treatment option for hepatic hydatid is crucial to ensure successful treatment outcome. Chemotherapy, newer approaches, like PAIR procedure, laparoscopy are available, but has limitations. Surgery is the mainstay of treatment. This study revealed, radical surgical excision coupled with chemotherapy and long-term aggressive chemotherapy for partially resected ensures lowest morbidity and mortality. Tailored surgical approach is key to successful outcome. Radical surgery is key to effective and successful outcome, ensures better quality of life and prevents recurrence of the disease. Decisive factor in low rates of recurrence is the cautious surgical technique.

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