

SURGICAL MANAGEMENT OF COMPLICATED LIVER HYDATID CYSTS IN ENDEMIC AREA THE REPUBLIC OF YEMEN

By

MARWAN MANSOUR BORHAM

Departments of General Surgery, Faculty of Medicine, Al-Azhar University, Nasr City,
Cairo, Egypt (*Correspondence: marwanborham1974@gmail.com)

Abstract

Hydatidosis is a zoonotic parasite caused by the larval of *Echinococcus granulosus* of worldwide distribution mainly sheep farming countries. Most of the complications are life-threatening and surgery is the best choice of treatment.

This study evaluated the clinical presentation and the best way to surgically manage complicated hydatid cysts in Yemen

This study was conducted in Alsalam Hospital in Saada Yemen Republic over two years From January 2017 to January 2019. The enrolled patients were 59 with hepatic hydatid cysts (38 females & 21 males), with mean ages of 32.5 years. The surgical procedures used according to their presentations were drainage in 25(42%), partial cystectomy and capitonnage in 15(25%), partial cystectomy and omentoplasty in 10(17%), per cystectomy in 5(8.3%), hepatectomy in 2(3.3%) and CBD exploration in 2 (3.3%).

The results showed that hepatic hydatidosis was a single cyst in 48 patients (81.3%) and multiple in 11 patients (18.6%). Patients were classified according to complications into wound sepsis in 4(6.7%), biliary leakage in 2(3.3%), intra-abdominal abscess in 3(5%), recurrence in 2(3.3%), bleeding in 2(3.3%) and lung atelectasis in 2(3.3%) Total postoperative morbidity was 25.4% and for surgical procedures and post-operative complication (P <0.05)

Key words: Yemen, Hydatid cysts, diagnosis, Surgery, Postoperative morbidity

Introduction

Liver hydatid disease (hydatidosis) was recognized since the Hippocrates time as liver full of water (Joshi *et al*, 2020). The life cycle of *Echinococcus granulosus* was first described by Haubner (1855), who experimentally infected a domestic pig with *E. granulosus* eggs dropped from dog and detected a fully developed hydatid cyst of liver (Kumaratilake and Thompson, 1982). Since then, echinococcosis/hydatidosis was reported in sheep rearing countries globally, with cycle sheep-dog-man (Haridy *et al*, 2000). This was particularly clear in South America, India, the Middle East, and Mediterranean Countries as well as worldwide by increasing travels and immigrations (Lopez-Marcano, 2017). In human, 50% to 75% of hydatid cysts were present in the liver (Eckert and Thompson, 1988), with about 72% of cysts in the right lobe with sizes varied from 1cm to 30cm (Armin *et al*, 2020). Hydatidosis apart from liver or lung unusual affected heart, kidney, bone and ocular (Gogus *et al*, 2003), CNS (Mazyad *et al*, 1998) or pancreas

(Akbulut *et al*, 2014) leading to significant morbidity and mortality (Gelman *et al*, 2000). Diagnosis of hydatid cyst started clinically and radiological imaging, ultrasound was the choice being easy, inexpensive clarified number, size, site, and cyst viability; also computerized tomography clarified its extent, depth and vascular invasion (Ghartimagar *et al*, 2013). Antibody assays can also add to radiological diagnosis, but rarely possible to exclude echinococcosis based on a negative serologic test (Sarkari and Rezaei, 2015).

There are three ways to treat hepatic hydatidosis; medical chemotherapy Albendazole[®] for three months, percutaneous aspiration of cyst and surgical intervention (Akcan *et al*, 2010). Surgical treatment in these cases is the must with completely elimination of cyst to avoid recurrence. The surgical treatments varied from complete resection (e.g., pericystectomy & hepatectomy), to minimal invasive procedures as percutaneous cystic aspiration (El-Sayed *et al*, 2020). But, there was an ongoing debate on the ideal treatment, as several factors affecting the choice of treat-

ment. For example, site and localization of cyst, experience of the operating surgeon, and presence of complications (Nunnari *et al*, 2012). Multiplicity of cyst is another common surgical problem, but sometimes management was difficult, requiring technical operative experience in this field (Prousalidis *et al*, 2004)

This prospective study was conducted to evaluate the long-term outcomes of various surgical techniques in treating large-sized, complicated hydatidosis in Yemen an endemic country and to compare post-operative complication, morbidity and recurrence.

Patient and Methods

This prospective study was conducted from January 2017 to January 2019, on 59 patients of both sexes and different ages. They were followed up for a period of two years in Department of Surgery; Al-Salam Hospital at Saada City, Yemen that's why there was delaying in publishing of this paper.

Preoperative evaluation: Patients underwent a detailed interview. Then, a thorough clinical examination was done as well as laboratory examinations for CBC, liver and kidney function tests, coagulation profile, ECG and chest X-ray, the ultrasonography was best tool in diagnosing hydatidosis. Computed tomography was indicated, which the ultrasound result was neither conclusive nor equivocal. The most important indication of computed tomography was cyst anatomical details; cyst multiple, solid components in cyst, intra-biliary rupture and/or cystic infection. Preoperative endoscopic retrograde cholangiography (ERCP) was done in case of suspected intra-biliary rupture or biliary tree obstruction based on biochemical, radiological or clinical evidence. If there was a suspicion of infection preoperative broad spectrum antibiotics was given, all patients with extra hepatic cysts were excluded.

Surgical technique: After confirmed diagnosis, patients were prepared for surgery. First of all patient was scrubbed. Then one of following incisions, right subcostal, bilateral subcostal, Mercedes Benzes, midline in-

cision was done according to the size and sites of cysts. In all hepatic hydatidosis patients, the area surrounding the cyst was covered with packs soaked with 0.5ml concentrated saline to prevent spread of the hydatid sands while the evacuating the cyst. The cyst contents were aspirated from a suitable surface using a needle, after aspiration 0.5ml concentrated saline solution was injected to sterilize the contents, then 1-2cm incision at the needle site to evacuate the whole contents. The germative membranes and daughter cysts were also removed using forceps or spoons. Besides, the redundant parts of the cyst were removed to allow better exposure. After the cyst evacuation, the cavity was irrigated using 0.5ml saline solution for 10-15 minutes. The cavity was then filled with warm saline, and was inspected for any possible bile leakage. After that, it was packed using dry white packs. Examination for bile leakage bile stains indicated bile communications, which was managed by a non-absorbable suture. Abdominal cavity was irrigated with 0.5ml saline solution in perforated case. Management of residual was done by a suitable technique based on the site, size and number of cysts as well as surgeon's preference. Techniques were: 1- Partial cystectomy and drainage, capsulorrhaphy, 2- Partial cystectomy and capitonage, 3- Partial cystectomy and omentoplasty, 4- Total cystectomy (per cystectomy), & 5- Partial hepatectomy.

In the first technique, cyst drainage was done after excision of superficial part with capsulorrhaphy by absorbable sutures around the cavity edges, and in the second one, dead space was obliterated by serious of purse strings sutures, starting from the bottom of per cyst. In the third one, flap of omentum was brought to rest within per cyst, and cavity was sealed small biliary leakage and obliterated the cavity. In the fourth one, excision of the endocyst and exocyst leaving raw area in the liver tissue, In the fifth one, excision of the cyst wall and part of the liver tissue if the cyst was concise to specific quadrant of the liver and peripherally located away from

portal vein and hepatic artery. If pre-operative evaluation indicated probability of connection between biliary duct and cyst, or suspected during surgery cholangiogram was done and placement of t tube.

Post-operative follow up all patients were given 3 courses of albendazole[®] of 4 weeks each with one week rest in-between and followed up as twice a month for three months then once a month for one year, and then every three months for another year.

Results

Patients were 59, of whom 38 were females (64.4%), and 21 were males (35.6%), with ages ranged between (15 & 50) years. Many patients were dog/cats owners or sheep breeders. The hepatic cysts were single in 48(81.3%), multiple in 11(18.6%), localization in right lobe in 39(66%), in left lobe 13(22%), and in both lobes in 7(11.8%). Patients were from rural area 42(72%) or urban ones 17(28%). The most symptoms was right hypo-chonderium pain in 45(76.6%), diagnosed on physical examination in 32 patients (54.2%), fever was in 18(30.5%), jaundice in 5(8.45%), nausea and vomiting in 40 (67.7%), itching in 5(8.4%), abdominal tenderness in 22(37.2%), abdominal distention in 1(1.6%), hepatomegaly with abdominal distention in 26(44%). They were classified according to their complications into infectious in 32(55.9%), interabiliary rupture in 20 (33.8%), and intera-peritoneal rupture in 7 (11.8%). Surgical outcome was extended right subcostal incision in 36(61%), bilateral subcostal incision in 15(25%), Mercedes Benz incision in 2(3.3%), and midline incision in 6(10%).

Surgical procedures were partial cystectomy & drainage in 25(42%), partial cyste-

ctomy & capitonage in 15(25%), partial cystectomy & omentoplasty in 10(17%), per cystectomy in 5(8.3%) hepatectomy in 2 (3.3%) and CBD exploration in 2 (3.3%). Patients 6(10.1%) had communication with biliary system either via bile duct branches or open in cavity or communications of cyst with common bile duct and obstruction of CBD from compression or infiltration by daughter cysts or hydatid fluids. Two (3.3%) underwent CBD explorations and insertion of T tube in CBD without perioperative complications. Others with biliary leakage into cyst (cystobiliary communication) were sutured with absorbable sutures and covered by omentum to seal non-visible small bile leak.

Post-operative complications: Post-operative morbidity was 25.4%, with wound sepsis developed in 4(6.7%) and the commonest early post-operative complications biliary fistula in 2 (3.3%) one in partial cystectomy with capitonage and the second in hepatic-tomy. All post-operative leakage healed by itself after conservative management without any surgical intervention. Intera-abdominal abscess was in 3(5%) one with partial cystectomy and capitonage the second in partial cystectomy with drainage, and partial cystectomy with omenplasty, bleeding in 2 (3.3%) one in hepatectomy and the second in partial cystectomy and capitonage, due to bleeding from cyst recurrence in 2 (3.3%) due to daughter cysts spillage and fluids into peritoneum cavity were in 2 (3.3%) due to iatrogenic adhesion or injury of diaphragm, without significant differences in incidence of early post-operative complications among different surgical procedures (P< 0.05).

Details were given in tables (1, 2, 3, & 4) and figures (1, 2, 3, 4, 5, & 6).

Table 1: Symptoms and presentation

Symptomatology	Number of patients	Percentage
Pain	45	45.27%
Abdominal swelling	32	54.23%
Fever	18	30.5%
Nausea ,vomiting	40	67.7%
Itching	5	8.4%
Abdominal tenderness	22	37.3%
Hepatomegaly	26	44%
Abdominal distention	1	1.6%

Table 2: Topographic distribution of cysts in liver

Age	15 up to 50 years old	
Sex	21 males	38 females

Table 3: Distribution of cyst in liver

Site	Number	Percentage
Single cyst	48	81.3%
Multiple cysts	12	18.6%
Right lobe cyst	39	66%
Left lobe cyst	13	23.7%
Bilobar cysts	6	10.1
Rural	42	56.1%
Urban	17	43.8%

Table 4: Surgical operation (n=59)

Surgical procedures	Patients No.	Percentage
Partial cystectomy and drainage	25	42%
Partial cystectomy and capitonage	15	25%
Partial cystectomy and omentoplasty	10	17%
Total cystectomy	5	8%
Partial hepatectomy	2	4%
Common bile duct exploration	2	4%

Table 5: Postoperative morbidity and complication (n=50)

Types of operation	Pcd	Pcc	Pco	Cystectomy + omentoplasty	Partial hepatectomy	Pe+cbd exp
Patient complications	25(42%)	15(25.4%)	10(16.9%)	5(8.4%)	2(3.3%)	2(3.3%)
Wound sepsis	2	--	--	1	1	--
Biliary leakage	--	1	--	--	--	1
Interabdominal abscess	1	1	1	--	--	--
Bleeding	--	1	--	--	--	1
Recurrence	1	1	--	--	--	--
Atelectasis	--	--	--	--	1	1
Total morbidity	25.4%					

Discussion

Patients with hydatid cyst should receive treatment follow up only in selected cases like calcified cyst (Greco *et al*, 2019), Albendazole® or Mebendazole® was effective in management in endemic area (Keong *et al*, 2018). The commonest complication was infection, cyst rupture and jaundice. (Arambulo *et al*, 1997) with surgery the main corner in management of hepatic hydatid cyst especially if non-operative management failed, and complicated disease (WHO, 2003) the site of hydatid cyst in the liver is considered important factor deciding the complexity of the cyst if it is in direct relation to big vessels like inferior vena cava, aorta, hepatic vessels or portal vein. Hydatid cysts of the liver most cases were asymptomatic 38 to 60% (Porcu *et al*, 2020).but in the present study, all showed clinical manifestations. The most common symptoms pain in the right upper hypochondrium and the right hypochondrial swelling. This agreed with Greco *et al*. (2019). Ultrasonography was

the most sensitive tools with specificity and sensitivity of 90% to 98% respectively.

In the present study, diagnosis depended on ultrasonography, confirmed at surgery with specific indications for CAT scan as anatomical cystic details, relation to other important structures, obstructive jaundice and solid component (Marom *et al*, 2019). The sensitivity of CT was 100% (Eyüboğlu *et al*, 2019). About one third of liver hydatidosis showed complications, mainly infection due to cyst rupture into biliary system, and less common intra-peritoneal or into viscous and vascular system rupture led to portal hypertension (Waghlikar *et al*, 2002). In the present study, infections were in 32 (54.2%), intera-biliary rupture in 20(33.8%), and intra-peritoneal rupture in 7(11.8%), and microbiological culture showed *Escherichia coli*. In the infected liver hydatidosis, stained with pile was accompanied by leakage into biliary trees. When these cysts rupture inside biliary canals, pyogenic organisms invade in as a secondary infection.

In the present study, intra-biliary rupture came as second common complications in 20(33.8%) patients. But, Waghlikar *et al.* (2002) in Turkey found that surgical procedures with complicated cysts were-infected: omentoplasty (2) and external drainage (3); intrabiliary rupture: omentoplasty (2) and internal drainage (2), and that postoperative morbidity was in 14 patients and was more in complicated cysts (6/11; 55%) compared to uncomplicated cysts (8/32; 25%)

Hepatic hydatidosis is therefore initially treated by surgical intervention. In this case, the surgery mainly aims at eradicating the parasite, preventing intra-operative spread of cyst contents and obliterating the residual cavity. The goal was achieved using different surgical procedures. First of all, scolicalid agents inactivated scoleces in cyst. In this study, 0.5% concentrated saline was introduced into cystic cavity for 10 to 15 for 15 minutes. Special precautions were taken in cases where the cyst and the biliary tract were connected together as the agent in such patients could lead to sclerosing cholangitis. In patients with relatively small-sized, sub-capsular cysts, non-anatomic hepatic resection was done. While in larger cysts affecting most of hepatic segments, anatomic resection was the technique of choice.

There are different surgical procedures for hydatidosis, all aimed to shorten obliteration period by performing proper internal and external drainage and prevent the possible accumulation of fluids in the residual cyst cavity. Among all these drainage, omentoplasty or captonnage with partial cystectomy were the done in this study. In liver hydatidosis patients with biliary tract involvement, it was necessary to explore biliary tract, perform a choledochotomy and place a T tube. Ductus choledochus exploration was done in two complicated patients with cyst rupture into the biliary tract. Routine pre-operative endoscopic retrograde cholangiopancreatography can alternate approach in similar cases. The common bile duct was cleaned of any germinal vesicles and thus fa-

cilitated drained (Dumas *et al.*, 1999) But, this technique was not routinely used.

In the present study, no death cases were reported during or after operation. Post-operative complications were 27%, but without significant between surgical procedures and complications as the basic surgical principles were thoroughly followed.

Conclusion

Hydatid cyst may be life-threatening if not early diagnosed and probably treated. Surgical intervention was considered the corner stone in treating complicated cysts; and although several surgical procedures were available, but without significant difference in outcomes of different techniques.

References

- Abuoglu, HH, Günay, E, Akyuz, C, et al, 2019:** Surgical approach to giant hepatic hydatid cysts: A single-center experience from Istanbul. *Int. J. Clin. Exp. Med.* 12:915-22.
- Akbulut, S, Yavuz, R, Sogutcu, N, Kaya, B, Hatipoglu, S, et al, 2014:** Hydatid cyst of the pancreas: Report of an undiagnosed case of pancreatic hydatid cyst and brief literature review. *World J. Gastrointest. Surg.* 6, 10:190-200
- Akcan, A, Sozuer, E, Akyildiz, H, Ozturk, A, Atalay, A, et al, 2010:** Predisposing factors and surgical outcome of complicated liver hydatid cysts. *World J. Gastroenterol.* 16, 24:3040-8
- Altıntaş, N, 1998:** Cystic and alveolar echinococcosis in Turkey. *Ann. Trop. Med. Parasitol.* 92:637-42
- Arambulo, P, 1997:** Public health importance of cystic echinococcosis in Latin America. *Acta Trop.* 67:113-24.
- Armin, A, Bizhan, Z, Amirhossein, E, Reza, S, Keivan, R, 2020:** Success fragment of a huge pulmonary hydatid cyst with lung-preserving surgery. *Case Rep. Surg.* 9526406. <https://doi.org/10.1155/2020/9526406>
- Dumas R, Le Gall P, Hastier P, et al, 1999:** The role of retrogradecholangiopancreatography in the management of hepatic hydatid disease. *Endoscopy* 31:242-71
- Eckert, J, Thompson, R, 1988:** *Echinococcus* strains in Europe: A review. *Trop. Med. Parasitol.* 39:1-8
- El-Sayed, YA, Ibrahim, EA, Morsy, TA, 2020:** Peri-cystectomy versus cystotomy and enucleation of germinal layer by tissue spearing

technique for lung hydatid in Egypt. J. Egypt. Soc. Parasitol. 50, 3:494-503.

Eyüboğlu, TS, Gürsoy, TR, Aslan, AT, Pekcan, S, Budakoğlu, H, 2019: Ten-year follow-up of children with hydatid cysts. Turk. Pediatri. Ars. 54, 3:173-8.

Gelman, R, Brook, G, Green, J, et al, 2000: Minimal change glomerulonephritis associated with hydatid disease. Clin. Nephrol. 53:152-8.

Georgiou, GK, Lianos, GD, Lazaros, A, Harysis, HV, Mangano, A, et al, 2015: Surgical management of hydatid liver disease. Int. J. Surg. 20:118-22.

Ghartimagar, A, Ghosh, M, Shrestha, O, Talwar, P, Sathian, B. 2013: 14 years hospital based study on clinical and morphological spectrum of hydatid disease. J. Nepal Med. Assoc. 52, 190:349-53.

Greco S, Canella R, Giambell, D, et al, 2019: Complications of hepatic echinococcosis: Multimodality imaging approach. Insights Imag. 10: 113-8.

Haridy, FM, Ibrahim, BB, Morsy, TA, 2000: Sheep-dog-man: The risk zoonotic cycle in hydatidosis. J. Egypt. Soc. Parasitol. 30, 2:423-9.

Joshi, D, Chowdhury, AT, Shrestha, MB, Al Mahmud, H, Salekin, S, et al, 2020: Complicated bilateral pulmonary hydatid cysts. J. Cardiothorac. Surg. Ther. 4:52-6.

Keong, B, Wilkie, B, Sutherland, T, et al, 2018: Hepatic cystic echinococcosis in Australia: An update on diagnosis and management. ANZ J. Surg. 88:26-31.

Kumaratilake, LM, Thompson, RC, 1982: A review of the taxonomy and speciation of the genus *Echinococcus* Rudolphi 1801. Z. Para

sitenkd. 68, 2:121-46.

Lopez-Marcano, AJ, Ramia, JM, Arteaga, V, et al, 2017: Percutaneous drainage as a first therapeutic step prior to surgery in liver hydatid cyst abscess: Is it worth? Wld. J. Hepatol. 9:114-8.

Marom, G, Khoury, T, Gazla, SA, et al, 2019: Operative treatment of hepatic hydatid cysts: A single center experience. Asian J. Surg. 42:702-7

Mazyad, MAM, Mostafa, MM, Morsy, TA, 1998: Spinal cord hydatid cysts in Egypt. J. Egypt. Soc. Parasitol. 28, 3:655-8.

Nunnari, G, Pinzone, MR, Gruttadauria, S, et al, 2012: Hepatic echinococcosis: Clinical and therapeutic aspects. Wld. J. Gastroenterol. 18: 1448-58

Porcu, A, Fancellu, A, Cherchi, G, Tsoulfas, G, Hoballah, J, et al, 2020: The role of emergency surgery in hydatid liver disease. In: Surgical Management of Parasitic Diseases, Springer, Cham.

Prousalidis, J, Kosmidis, CH, Fahantidis, E, Harlaftis, N, Aletras, O, 2004: Surgical treatment of multiple cystic echinococcosis. HPB (Oxford). 6, 2: 110-4.

Sarkari, B, Rezaei, Z, 2015: Immunodiagnosis of human hydatid disease: Where do we stand? Wld. J. Methodol. 5, 4:185-95.

Waghlikar, GD, Sikora, SS, Kumar, A, Saxena, R, Kapoor, VK, 2002: Surgical management of complicated hydatid cysts of the liver. Trop. Gastroenterol. 23:35-7

WHO, 2003: Informal Working Group International classification of ultrasound images in cystic echinococcosis for application in clinical and field epidemiological settings. Wld. Hlth. Org. Geneva, Switzerland.

Explanation of figures

F. 1: Complicated cyst related to inferior vena cava, F. 2: Complicated cyst with cyst biliary connection, F. 3: Closure of cyst biliary connection with absorbable suture, F. 4: Evacuation of huge hydatid cyst, F. 5: Complete excision of the cyst, F. 6: Partial hepatectomy of left lobe

