

**Basrah Journal****of Surgery****Bas J Surg, September, 12, 2006****UNUSUAL PRESENTATION OF A COMMON DISEASE****Mazin A Abdullah\*, Noori H Jasim#, Furat Shani@**

\*CABS, Lecturer, Dept of Surgery, College of Medicine, University of Basrah. #FICMS, Lecturer, Dept of Surgery, College of Medicine, University of Basrah, @CABS, Lecturer, Dept of Surgery, College of Medicine, University of Basrah

**Abstract**

This study aimed to describe the clinical features of acute rupture of hydatid cyst in the peritoneum. We retrospectively studied 7 patients who underwent explorative laparotomy between January 2002 and October 2005, and in whom exploration confirmed the rupture of hydatid cyst in the peritoneum. Rupture was secondary to trauma in 2 patients, and occurred spontaneously in 5 patients.

All patients presented with acute peritonitis, and anaphylactic shock developed in 4 patients. One patient died postoperatively. Ultrasonographic diagnosis was made in 3 patients. Surgical treatment consisted of the treatment of peritonitis, the ruptured hydatid cyst and evacuation of the free intraperitoneal hydatid fluid. All patients kept on albendazole. Acute rupture of hydatid cyst in the peritoneum should be considered when evaluating acute abdomen in endemic areas.

**Introduction**

**H**ydatic disease (Echinococcosis) is a common disease distributed in most of the world, especially in the sheep rearing areas where it is endemic in many countries including Iraq<sup>1-4</sup>.

The disease can be caused by one of three species of the genus Echinococcus (E. granulosus, E. multilocularis & E. voegeli)<sup>5,6</sup>.

Echinococcal cyst is the larval cystic stage of a small taeniid type tape worm (usually E. granulosus)<sup>7,8</sup>.

The adult worm (3-6mm long) inhabits the small intestine of carnivorous definitive hosts such as dogs or wolves, and echinococcal cyst stage occur in herbivorous intermediate hosts such as sheep, cattle and goats<sup>7,9</sup>.

In the typical dog-sheep cycle, tape worm eggs are passed in feces of an infected dog and may subsequently be ingested by sheep, they hatch into

embryos in the intestine, penetrate the intestinal lining, and then are picked up and carried by blood throughout the body to major filtering organ (mainly liver and, or lungs). Then they transform and develop into larval echinococcal cysts in which numerous heads (protoscolices) are produced by asexual reproduction<sup>1,2,7,10,11</sup>. Cysts are produced by the peri parasitic host tissues (pericyst) which encompasses the endocyst of larval origin, inside the laminated layer the cyst is covered by a multipotential germinal layer giving rise to bulb-like processes which are termed brood capsules which produce numerous protoscolices.<sup>6</sup> In addition daughter cysts of variable size are often detected. The clinical features of hydatid disease are highly variable<sup>1,2</sup>. Cystic lesions cause symptoms via compressing adjacent organs or may be

totally silent. Sudden symptomatology is usually due to spontaneous or traumatic rupture of the echinococcal cyst with or without anaphylaxis<sup>12,13</sup>. Cysts may rupture into the peritoneal or pleural cavity<sup>12-16</sup> bile ducts, the pericardium or even blood vessels leading to extraordinary manifestations and severe complications. This article discusses those patients who presented as acute abdomen due to rupture of the cyst.

### Methods

We retrospectively studied 7 patients presented with features of acute abdomen, and who had underwent surgery between January 2002 and October 2005 in Basra General Hospital and in whom the exploration had confirmed the diagnosis of acute rupture of echinococcal cyst in the peritoneum. We present the cases shortly.

### Case 1

A thirty years old man presented with generalized abdominal pain few hours after a road traffic accident. His blood pressure was normal but he had tachycardia, the abdomen was tender all over with rigidity. No sonographic or x-rays service was available, so explorative laparotomy was decided. On exploration, huge amount of bile stained fluid was found in the peritoneal cavity and a ruptured large hydatid cyst found in the right-lobe of liver, which was still containing some daughter cysts, no other pathologies found. After clearing of the cyst, suction of free fluid and irrigation by hypertonic saline, abdomen closed leaving a tube drain.

The drainage of bile stained fluid continue for one month of about 500 ml. daily. The patient referred for ERCP in a gastro-entrolgy center

where a filling defect found in the common bile duct (membrane of the cysts).Sphincterotomy performed and the drainage ceased after two weeks. Patient was kept on albendazole.

### Case 2

A thirty two years old man was admitted to the waiting surgical ward because of sudden, severe upper abdominal pain of 2 hours duration. There was no history of trauma; patient rapidly passed into a shock state, pulse was 130/minute. The abdomen was rigid and mildly distended.

Resuscitation started by intravenous ringer solution in addition to hydrocortisone. After improvement sonographic examination (figure-1) discovered a ruptured hydatid cyst in the left lobe of the liver, the cyst found collapsed but no free intraperitoneal fluid could be detected. The cyst dealt with surgically in the routine way. Patient improved gradually and discharged well after 8 days.

### Case 3

A fifty years old man presented with shock to the medical emergency unit. His pulse rate was 120/ minute and his blood pressure was 80/40 mm. Hg, his ECG was normal apart from tachycardia. Chest was clear. Resuscitation started and surgical consultation Sought.

The surgeon found tense, tender abdomen, and sonographic examination revealed presence of free intrapeitoneal fluid. Chest x-ray was normal. Emergency explorative laparotomy confirmed presence of a ruptured hydatid cyst in right lobe of liver. Abdomen closed after dealing with the cyst and leaving a drain. Patient discharged well after 10 days and was kept on albendazole.

**Case 4**

A 46 years old lady with history of trauma presented with acute abdomen. She got tachycardia while her blood pressure normal. Ultrasound examination found multiple cysts free in the peritoneal cavity while chest x ray & ECG were normal.

Explorative laparotomy performed few hours later and revealed a ruptured large hydatid cyst in the right lobe of the liver with spillage of hundreds of daughter cysts of varying sizes all over the peritoneal cavity. All of which removed and the hepatic cyst cleaned.

Patient passed a smooth postoperative period and discharged well after 12 days on albendazole tablet.

**Case 5**

A 35 years old man presented to the emergency unit because of sudden central abdominal pain. There was no history of trauma.

His examination revealed pulse rate of 110/ minute blood pressure 100/80 mm.Hg. Sonographic examination showed a large cyst in right lobe of the liver with presence of daughter cysts and small amount of free intraperitoneal fluid. Chest x ray was normal. Laparotomy confirmed the rupture of hydatid cyst of the liver, which treated by endocystectomy and removal of all daughter cysts. Patient received hydrocortisone intraoperatively and a tube drain left. He was discharged well after 6 days.

**Case 6**

A 55 years old woman brought in shock state to the emergency unit; she was complaining of sudden abdominal pain and vomiting. Resuscitation started and abdominal examination revealed a distended, tender abdomen. Sonographic examination done which showed presence of large amount of

free intraperitoneal fluid and chest x ray was normal. Explorative laparotomy performed, abdomen was filled with hundreds of small hydatid cysts, with ruptured cysts found in liver and in mesentery of small bowel. Patient received hydrocortisone pre and postoperatively and despite the resuscitating measures patient's condition continues to deteriorate and finally patient died shortly after operation.

**Case 7**

A 13 years old boy was admitted to the emergency surgical unit because of a generalized abdominal pain, that occurred after falling on the ground while playing with his friends one day before admission.

The pain was dull in nature and after few hours it was localized in right iliac fossa and became more severe the patient got also nausea, anorexia with low grade fever. The abdomen was soft, with tenderness in right iliac fossa & positive rebound tenderness. There was leucocytosis and his urine exam was normal. Ultrasound examination was not available.

Provisional diagnosis of acute appendicitis was reached, and because of history of trauma, the abdomen opened through lower mid line incision. Appendix found normal, and there was a large amount of intraperitoneal fluid, so the incision extended upward and a ruptured cyst of the spleen found which was involving most of the spleen so splenectomy was performed. Abdomen closed with drain. Patient discharged well after 10 days on albendazole and monthly benzathine penicillin. His Chest x ray postoperatively was normal.

**Results**

Over The period of the study (about 3

years) 69 patients had undergone surgery for hydatid cyst in the abdomen in Basra General Hospital, 7 of who presented as acute rupture of the hydatid cyst in the peritoneum that is to say 10.14% of the cases. Rupture of the cyst was secondary to trauma in 2 patients and occurred spontaneously in 5 patients. Diagnosis was made preoperatively in 3 patients by the aid of ultrasonographic examination. Anaphylactic shock was present in 4 patients. Five patients had the cyst in the liver, one patient had the cyst in the liver & mesentery & one patient had the cyst in the spleen. No lung hydatid found in these patients. Summary of the cases seen in the table I.

### Discussion

Hydatid cyst enlarge gradually and slowly<sup>11</sup>, eventually complications ensue in particular rupture of the hydatid cyst in the peritoneal cavity, where it may cause a hypersensitivity reactions, varying from urticaria to anaphylactic shock<sup>17,18</sup>.

In the period of this study (34 months), 69 patients had undergone surgery for hydatid disease of the abdomen in Basrah General Hospital, 7 of them presented as acute abdomen that is 10.14% of all cases which is similar to some reports<sup>2,4</sup>.

The incidence of cyst rupture is variable in different studies from 1.75 to 10%<sup>5,19</sup>. Rupture of hydatid cyst into the peritoneal cavity causes peritonitis with anaphylactic response, shock and dyspnea<sup>20,21</sup>. The degree of hypersensitivity depends on dose of spillage and condition of body immune system<sup>20</sup>. Our patients developed features of acute abdomen due to peritonitis and 4 of them had unexplained shock which later was attributed to anaphylaxis, and there are reports of anaphylactic shock being the

only clinical manifestation of the hepatic hydatid cyst<sup>22</sup>. The incidence of anaphylaxis in hepatic hydatid disease varies and was a result of cyst perforation generally occur on needle aspiration or open surgery; however the spillage of cyst fluid with intravascular spread resulting from trauma may also trigger anaphylaxis<sup>12,13,23</sup>. Rupture of hydatid cyst caused either by trauma ranging from minor trauma to a significant blunt trauma, it may occur spontaneously, or even after cough episode<sup>14,24,25</sup>.

In our patients there was history of significant blunt trauma (case 2) and mild trauma (case 7), while the cyst ruptured spontaneously in the remaining cases.

Ultrasonography is the investigation of choice for diagnosis of asymptomatic echinococcal cyst<sup>3,11,26-29</sup>. In our patients preoperative diagnosis made by help of ultrasound examination in 3 of 5 patients, and it was unavailable in 2 patients.

Liver is most commonly involved by the cyst (55-70%), followed by the lung (18-35%), and two organs can be affected simultaneously in about 5-13% of cases<sup>30,31</sup>. Liver found involved in 5 of our patients, in the 6th patient cyst found in the liver and mesentery at same time. While it was in the spleen in the last patient. All patients showed features of acute abdomen for which explorative laparotomy was done.

The management in such cases depends on surgical intervention, in addition to IV hydrocortisone in a dose of 500-1000 mg with subcutaneous adrenaline (1 ml of 1/1000 solution) or anti-histamine<sup>32</sup>.

Surgical intervention consists of dealing with the ruptured cyst by endocystectomy, evacuate the free fluid, removal of daughter cysts, liver cavity management, in addition the

peritoneal cavity should be flooded with liberal quantities of a scolicide followed by aspiration and then irrigation with saline to wash out the remaining scolicide agent followed by adequate drainage.

One patient (case 1) needed ERCP because of continuous drainage of bile for more than 4 weeks, so endoscopic sphincterotomy performed and exterior drainage stopped. In this patient both intraperitoneal and intrabiliary rupture had occurred similar cases has been reported in the literatures. There is an increase risk of septicemia after splenectomy for any indication so splenectomy should if possible be avoided 8. However in case 7, splenectomy was performed as the cyst was occupying most of the spleen.

Although mortality directly due to

hydatid disease is very low, we lost one patient (case6) as this patient develop anaphylactic shock which didn't respond to all resuscitative measures. A mortality rate between 0.29-0.6percent has been reported<sup>33</sup>.

In our series Medical treatment should follow surgical treatment<sup>4,13</sup>.

All our patients were kept on albendazole, in a dose of 400 mg bid with meal for 28 days; we repeat a cycle of 28 days after stopping treatment for 14 days to a total of 3 cycles<sup>3, 5</sup>. Unfortunately our patients were difficult to be followed our follow up ranging from 2 weeks-6months.

In conclusion Rupture of hydatid cyst intraperitoneally should be considered in the differential diagnosis of acute abdomen in infested area.



**Figure 1: Normal liver with transonicCyst (11 by 6.7 cm) with thick internal echogenic bands (ruptured complicated hydatid cyst!)**

**Table 1: Summary of the cases**

No of case	Age in years	Sex	Presence of shock	History of trauma	Site of the cyst	Outcome	u/s diagnosis
1	30	M	-ve	+ve	Liver	Improved	unavailable
2	32	M	+ve	-ve	Liver	Improved	+ve
3	50	M	+ve	-ve	Liver	Improved	-ve
4	46	F	-ve	-ve	Liver	Improved	+ve
5	35	M	+ve	-ve	Liver	Improved	+ve
6	55	F	+ve	-ve	Liver&mesentry	Died	-ve
7	13	M	-ve	+ve	Spleen	Improved	unavailable

## References

- 1- Aly M. Rady: Hydatidosis ancient,widespread harmful disease. Postgraduate Doctor-middle east. 1980 Volume; 330-36.
- 2- Siraj S. Malaika; Ahmad Attayeb ; Saleh Sulaimani; Jakkidi J. Reddy: Human Echinococcosis in Saudi Arabia. Saudi Medical Journal, April 1981; 2(2): 77-84 .
- 3- Mc Manus DP, Z hang W, Li J ,Bartley PB: Echinococcosis. Lancet 2003,18:362 ;1295-1304(Medline).
- 4- Moro P.,Gonzales A., Gilman R. Hunter's Tropical Medicine and Emerging disease. 8<sup>th</sup> Ed, Philadelphia: W.B. Saunders.2000 {Medline}
- 5- King C, G. Mandell, J. B ennett, and R. Doline: Principles &Practice of Infectious Diseases .5<sup>th</sup> Editors .New York, Churchill Livingstone. 633-40.
- 6- G.M. Edington, H.M. Gilles; Pathology in the Tropics.2<sup>nd</sup> ed. London, Edward Arnold, 1967.201-7.
- 7- Eckert J, Deplazes P: Biological, epidemiological, and clinical aspects of echinococcosis a zoonosis of increasing concern. Clin Microbiol Rev, 2004; 17(1):107-35(Medline).
- 8- As hour Gebreel : Hydatid disease. Postgraduate Doctor-Middle East 1983 Volume 3; 11-17.
- 9- Alexander J. Walt: Cysts and benign tumors of the liver. The Surgical Clinics of North America, Hepatic surgery, April 1977 Volume 2 ,449-64.
- 10- Raouf M. Sallam , Patrick A. Duffy , Abdul Hadi M. Oma: Hydatid Cyst of the Submandibular Salivary Gland: Case Report . Saudi Medical Journal. 1982,3:43-4.
- 11- Sayek J, Timaksiz MB, Dogau R: Cystic hydatid disease: current trends in diagnosis and management, Surg . Today.2004;34(12):987-96.
- 12- Buyuk, Yalen a, Turan, Arzu Akeay b, Uzun, Ibrahim b, Aybar, Yesim C, Cin, Ozgur b, Kurnaz, Gulay b: Non-ruptured hydatid cyst can lead to death by spread of cyst content into bloodstream: an autopsy case. European Journal of Gastroenterology and Hepatology, 2005,17(6):671-673.
- 13- Biyabani S.R., Abbas Farhat, Ghaffar Shehzad, Talati Jam: Unusual presentations of hydatid disease of the urinary tract, The Journal of Urology, 2000,163(3)869-76.
- 14- Pinar Polat , Mecit Kantarci, Fatih Alper: Hydatid disease from head to toe. Turkish Journal of Trauma and Emergency Surgery.2003(9):212-14.
- 15- Van Gulik TM, Lygidakis NJ, Vander Hyde MN: Intra biliary rupture of hydatid cyst of the liver with concomitant intraperitoneal rupture into lesser sac. Neth. J. Surg. 1987;39(6): 194-6.(Medline)
- 16- Navsaria PH, Forlee MV, Nicol AJ: Traumatic rupture of a hepatic cyst. S Afr J Surg. 2003;40(3):103-5.(Medline)
- 17- Rady AM: Splenic Echinococcosis. Journal of Kuwait Medical Association 1978;12:187-92.
- 18- Gabriel A Kune: Liver hydatid disease. Surgery middle east ed , I 981 ;2(2):77-84.
- 19- Beyrouiti MI, Beyrouiti R, Abbas J, Kharrat M, Ben Amar M, Frika F, Elleuch S, Gharbi W, Chaabouni M , Ghorbel A: Acute rupture of hydatid cysts in the peritoneum: 17 cases. Presse Med. 2004; 33 (6) 378-84.
- 20-Nelson A WORL , Primary Surgery ed. New York, Me GrawHill, 1979; 1415-1420.
- 21-Durif S, Marinkovic Z.Febvre C, Raffoul J: Hydatid liver disease revealed by surgical acute abdominal syndrome, Arch Pediatric.2005;12(11):1617-9.
- 22- Geogiou S , Maroulis J et al: Anaphylaxis shock as the only clinical manifestation of hepatic hydatid disease, Int. J. Dermatology, 2005; 44 (3) 233-7.(Medline).
- 23- Kantarci M, Onbas O, Alper F, Celebi Y. Yigiter M ,Okur A: Anaphylaxis due to a rupture of hydatid cyst imaging findings of a 10 yr old boy. Emerge Radiology 2003,10(1):49-52.
- 24- Ayhan Koyunca, Mustafa Turan: Traumatic pelvic hydatid cyst rupture; report of a case Turkish Journal of Trauma and Emergency Surgery.2003;9:213-14.
- 25- Navasaria PH, Forlere MV, Nicol AJ. Traumatic rupture of a hepatic hydatid cyst. S Afr J Surg, 2003; 40(3): 103-5(Medline).
- 26- Lee SH, Scudamore CH: Combined radio logic and endoscope management of biliary obstruction from ruptured hydatid disease. Gastrointestinal endoscopy, 1991; 37(1): 73-5.
- 27- Richter J, Hatz C, Haussinger D: U Ultrasound in tropical and parasitic disease, Lancet, 2003; 362 (9387). 900-2.
- 28- Sayek J, Onat D: Diagnosis and treatment of uncomplicated hydatid cyst of the liver, World J Surg, 2001; 25 (1): 21-7.
- 29- Akhmedov JC, Ultrasound examination in diagnosis of hydatid echinococcosis, Khirurgiia, 2004; 3: 18-22. {Medline}.
- 30- Kir A, Barun E: Simulatneous operation for hydatid cyst of right lung and liver , Thorac Cardiovas Surgeon, I 995;43: 62-4.
- 31- Guntz M, Coppo B, Lorimier G, Cronier P, Hydatid cyst of the liver appearing late (10-22 years) after surgical treatment of pulmonary hydatidosis, Physiopathologic problems J Chir Paris, 1990: 127: 375-81.
- 32- Nelson Awori: Primary Surgery 2<sup>nd</sup> ed . New York , Mc GrawHill , 1997 : 1415-1420.
- 33- Chen WQ: Surgical management of complicated hydatidosis Chung-Hosa-Wai-Ko-Tsa-Chih ,1992 ; 30:216-7.