

HYDATID DISEASE OF THE LIVER IN CHILDREN

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ABSTRACT:

Twenty one patients with hydatid disease of the liver were operated upon between 1986-1997. There were 10 males and 11 females. The mean age of the patients was 6.5 years (range 3 to 12 years). Abdominal distention with mass was the commonest method of presentation symptoms (71.4%). This was followed by abdominal pain (38%). Hepatomegaly with palpable mass was present in 57.1%. Three patients had concomitant pulmonary and brain hydatid disease. The diagnosis was made using clinical, skin testing, serology and imaging techniques. All patients received a preoperative course of Mebendazole (50mg/kg/day) for a period ranging between 1-8 weeks. At surgery, 11 patients (52.3%) had a single cyst and 8 were located in the right lobe of liver. Ten patients (47.6%) had multiple cysts occupying both lobes. Three forms of surgical treatment were used: Capitonnage + partial excision of fibrous capsule; total excision of the cyst; and external drainage of cyst cavity. We have to re-operate in 3 patients (14.2%). The mean follow up of patients was 24 months. Although there were no deaths in the series, 5 patients (23.8%) developed postoperative complications. Surgical treatment in the form of primary closure of the cyst cavity without drainage, seems to offer the best therapeutic option in these patients.

Key words: Liver cysts, hydatid disease, echinococcosis.

INTRODUCTION:

Hydatid disease is a major endemic health problem in certain areas of the world^(1,2). The most commonly affected organ is the liver^(1,2,3). Children are susceptible to this parasitic infection, and the concept that hydatid cysts are not seen in young patients because of a slow pathologic process is not valid. Medical treatment using benzinidazole compound is of limited value⁽¹⁾. Surgery is the treatment of choice in hepatic hydatid disease. We report our experience with hepatic hydatid disease in children between 1986-1997.

PATIENTS AND METHODS:

We reviewed the records of 21 consecutive paediatric patients admitted to the Maternity and Children's Hospital, King Faisal Specialist Hospital and King Khalid University Hospital in Riyadh between 1986-1997 with hydatid disease of the liver. We extracted data relating to age, sex, hospital stay, geographical area of origin, clinical patterns, investigations, treatment, postoperative morbidity and mortality, and the incidence of recurrence. In all cases the diagnosis was confirmed at laparotomy and by histopathological examination. All patients underwent surgical treatment.

RESULTS:

Twenty one patients were treated for hydatid liver cysts between 1986-1997. (10 males and 11 females). The mean age was 6.5 (range 3 years to 12 years). All patients were Saudi except three. Seven were from the central region of Saudi Arabia, 6 from the North and 5 from the South.

The overall hospital stay ranged from 11 to 42 days with a mean of 23 days. The mean hospital stay in patients with primary closure was 21.9 days versus 27.5 days in patients with non-primary closure. Patients had symptoms from 1 day to 1 year with an average duration of 5 months. The commonest presentation, abdominal distention with a mass, was present in 15 patients (71.4%). Recurrent abdominal pain was present in 8 children and vomiting in two. Two presented with upper abdominal pain for 1 day following blunt trauma to the abdomen; one had an infected hydatid cysts of the liver, and one had a ruptured cyst with anaphylactic shock.

Three patients had no abdominal symptoms; the presenting symptoms were respiratory in two and loss of vision in one; further investigation disclosed hepatic hydatid disease with concomitant pulmonary and brain cysts. In terms of physical findings, 12 patients (57.1%) had hepatomegaly with a palpable mass. Six patients had hepatomegaly only. None had fever, jaundice or signs of peritonitis.

Laboratory data: two patients had leucocytosis; eosinophilia was found in eight (38%). Liver functions tests were normal in all patients. A Casoni test was positive in four of eight patients (50%). Indirect haemagglutination (IHA) was positive in ten of eleven patients tested. (92%) (Table 1).

Imaging studies included plain abdominal films, ultrasound and CT scan of the abdomen. In one of ten patients plain films showed calcification in the right upper quadrant. Ultrasound of the abdomen was carried out in 20 patients and was diagnostic in all. CT scan of the abdomen was performed in 11 patients giving more detail about the

site, size, and number of the cysts (Fig. 1). Five patients had concomitant extrahepatic hydatid cysts; lungs were involved in three and brain in two.

Preoperatively, all patients received Mebendazole (50mg/kd/day) for a period ranging from one to eight weeks (average 2 weeks). In 16 patients, Mebendazole was continued postoperatively for 1-6 months.

All patients underwent surgical treatment. Laparotomy revealed a single cyst in 11 patients (52.3%). In eight, it was located in the right lobe of the liver. Ten patients (47.6%) had multiple cysts occupying both lobes. The cyst size ranged between 2 and 20 cm (average 12 cm). Twenty one primary surgical procedures were carried out. Cetramide 0.5 - 1% was used for intraoperative sterilization in most patients.

Three patients had total excision of the cyst, of whom 2 had a single lesion. Evacuation of the cyst contents and endocystectomy were used in the remaining seventeen. The residual cavity was managed in several ways (Table 2). In 12 patients (57.1%), the procedure was capitonnage (obliteration of residual cyst cavity with transfixion sutures from the depth to the surface) and partial excision of fibrous capsule. In five external drainage of the residual cavity was carried out. One patient had introflexion of the cyst cavity (obliteration of the cavity by folding the cyst wall inside the cavity and suturing the outer surface layers to the bottom of the cavity to fix the folded cyst wall). All specimens were submitted for histopathology which confirmed the diagnosis.

Postoperative complications were seen in 5 patients (23.8%). Two developed a biliary fistula which closed spontaneously. One patient had a biliary cyst, jaundice and a right sided pleural effusion. The cyst was treated by surgical drainage; the pleural effusion was managed conservatively. Another patient developed acute renal failure, plus a severe hepatic insult with raised liver enzymes and high bilirubin, and a right femoral artery false aneurysm at the site of the venous and arterial cannulation for hemodialysis. The renal failure was attributed to methyhemoglobinemia secondary to absorption of

silver nitrate used for intraoperative sterilization. This patient's renal and hepatic insults recovered completely in 6 weeks but he required emergency repair of a ruptured right femoral aneurysm. One patient developed adhesive bowel obstruction which required laparotomy and enterolysis. We had to re-operate in 3 other patients (14.2%), in two because of cysts missed during the first laparotomy and in one due to persistence of a residual cyst at the site of a previous excision. In the latter patient a biliary cyst was found and drainage was effected. There were no death. The mean follow-up was 24 months.

DISCUSSION:

Hydatid disease is a parasitic infection caused by the larval form of echinococcus granulosus. The commonest sites of infestation are the liver and lungs^(1,2,3). Hydatid disease is endemic in countries around the Mediterranean, Australia, South America and a few areas in North America and the U.K.⁽⁴⁾. The disease is not uncommon in Saudi Arabia. Although an epidemiological study has not been done, there appears to be an increasing incidence in this country. This may reflect better diagnostic facilities and improved medical care throughout the Kingdom. In endemic areas, the disease is more prevalent in rural communities and is usually acquired by living in contact with sheep and dogs⁽³⁾. All of our patients were from rural areas of the Kingdom and most lived in contact with sheep and dogs. Most children tend to be infected by contact with dogs, although some are indirectly infected by eating food contaminated with parasitic eggs⁽⁵⁾. Hydatid disease is found in all age groups with the highest incidence during the second, third and fourth decades of life⁽⁶⁾. Children, even very young ones, can be affected and may present with large hepatic cysts^(5,6,7,8). In endemic countries, up to 25% of hydatid disease is reported in children^(9,10). Our youngest patient was 3 years.

The commonest method of presentation in our series was abdominal distention with a mass (71.4%) followed by abdominal pain (38%). The most common physical findings were hepatomegaly with palpable mass (57.1%) followed by hepatomegaly (28.5%). None of our patients presented with fever, jaundice or signs of an acute abdomen. These findings are comparable with the clinical presentation reported in other children^(3,9). Abdominal pain and an abdominal mass, with or without hepatomegaly, is the commonest clinical presentation in adult patients^(11,12,13).

The preoperative diagnosis in this series was based on a combination of clinical, serological and radiological assessments. Eosinophilia was non-specific and was present in less than half of our patients. The Casoni test and IHA were the only tests available to us. The IHA was positive in 91% of patients tested. The Casoni test and IHA have been

reported to be positive in 50-90% and 70-86%, respectively^(2,3,4,5,6,9,12). Ultrasound and CT scan were helpful in all of our patients but the CT scan gave more information about the size, site and number of cysts.

Surgery is the only effective treatment for hepatic hydatid disease and is recommended for both symptomatic and asymptomatic patients^(1,4,12). Most surgeons will agree that proper management must include sterilization of the cysts, avoidance of spillage, evacuation of the cyst, and removal of the germinal layer. Numerous solutions have been used as scolicedal agents; however, some of these such as formalin and silver nitrate are toxic^(14,15). Cetrimide (0.5-1%) is commonly used with no major side effects; this was our choice. Only local toxicity causing chemical peritonitis has been reported⁽¹⁶⁾.

The management of the residual cavity is the most controversial aspect of surgical treatment as exemplified by the various procedures in this paper. In essence, cyst can be evacuated by the use of external tubes, by marsupialization or by internal drainage; alternatively they may be closed primarily without drainage using saline instillation, capitonnage or omentoplasty^(3,11).

The majority of our patients had closure of the cyst cavity without drainage; this was associated with fewer complications and a shorter hospital stay. Demicri et al⁽¹¹⁾ and Sayek et al⁽¹⁷⁾ found that external drainage significantly increased postoperative morbidity and was associated with a longer hospital stay.

Total excision should be reserved for peripherally placed cysts, pedunculated lesions, and extrahepatic intra-abdominal cysts, although major hepatic resection has been advocated⁽¹⁸⁾.

Reported postoperative complications include wound infection, hemorrhage, anaphylactic shock, peritonitis, subphrenic abscess and external biliary fistula⁽²⁾. In this series postoperative complications occurred in 23.8% which is comparable to other studies^(1,19). Recurrence rates varying from 0.5% to 22% have been reported (average 5-10%)^(5,12,13,20,21).

We had to reoperate on 3 patients (14%). Two cysts were missed during the first laparotomy which was for multiple hydatid cysts; one cyst resulted from an accumulation of bile in the residual cavity.

Morbidity and mortality have decreased during the last 20 years (0-4%)^(2,4,21). None of our patients died. The efficacy of benzimidazole compounds, mebendazole or albendazole, in the treatment of established hepatic hydatid disease remains uncertain^(12,21). Mebendazole or albendazole have been claimed to be effective in high doses^(19,22,23,24). We like others, used this medication hoping to prevent recurrence in case of accidental intraperitoneal spillage of cyst contents. Fortunately, we did not have extrahepatic intra-abdominal cyst formation secondary to intraoperative spillage.

In conclusion, hepatic hydatid disease is not uncommon in children and can present in patients in an advanced state. An accurate diagnosis can be made by a combination of clinical, serological, and radiological examinations. At present, surgery seems to offer the best therapeutic option. In our opinion, primary closure of the cyst cavity without drainage is best for patients with uncomplicated hepatic hydatid disease because it is associated with a shorter hospital stay and a low complication rate. Health education in endemic areas will probably decrease the incidence of this parasitic infection in the future.

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**TABLE I: LABORATORY, SEROLOGICAL AND
RADIOLOGICAL DATA**

| TEST | NO. OF PATIENTS TESTED | NO. OF ABNORMAL RESULTS |
|-------------------------------------|-----------------------------------|--|
| Liver function | 21 | 0 |
| White blood count | 21 | 2 |
| Eosinophils | 21 | 8 |
| Casoni | 8 | 4 |
| Indirect haemagglutination (IHA) | 11 | 10 |
| Plain abdominal x-ray | 10 | 1 |
| Ultrasound abdomen | 20 | 20 |
| CT scan abdomen | 11 | 11 |

**TABLE II: PRIMARY SURGICAL
PROCEDURES AND COMPLICATIONS**

| SURGICAL PROCEDURE | NO. | COMPLICATIONS | NO. |
|---|------------|---------------------------------|------------|
| Capitonnage + partial excision of fibrous capsule | 12 | Biliary fistula | 1 |
| | | Biliary cysts } | |
| | | Jaundice } | 1 |
| | | Pleural effusion} | |
| Total excision | 3 | Adhesive bowel obstruction | 1 |
| External drainage of residual cavity | 5 | Biliary fistula | 1 |
| | | Acute hepatic failure } | |
| | | Acute renal failure } | 1 |
| | | False femoral artery aneurysm } | |
| Introflexion | 1 | --- | --- |
| TOTAL | 21 | | 5 |

LEGENDS:

FIG. 1: Abdominal computed tomographic scan demonstrating multiple hepatic hydatid cysts in a 7-year-old child.

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