



## Hydatid Cysts in Children

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**Background/Purpose:** Hydatid disease is a parasitic infection caused by a parasite, *echinococcus granulosus*, characterized by cystic lesion in the liver, lungs and rarely in other parts of the body. Yemen is a highly endemic area for hydatid disease, especially in the northern areas with sheep raising. Prevention is the main method for eradication but still having its limitations. Surgery is the cornerstone of management of pulmonary hydatid cysts and aims to remove hydatid cysts or its remnants and obliterate the residual cavity. The aim of this study is to identify the optimum and safe way for treating children having pulmonary hydatid cysts and living in the highly endemic areas with the least possible complications. This study included characteristics on presentation, investigation and operative techniques and postoperative morbidity and mortality in a group of patients with pulmonary hydatid cysts.

**Materials & Methods:** Hydatid disease is a parasitic infection caused by a parasite, *echinococcus granulosus*, characterized by cystic lesion in the liver, lungs and rarely in other parts of the body. Yemen is a highly endemic area for hydatid disease, especially in the northern areas with sheep raising. Prevention is the main method for eradication but still having its limitations. Surgery is the cornerstone of management of pulmonary hydatid cysts and aims to remove hydatid cysts or its remnants and obliterate the residual cavity. The aim of this study is to identify the optimum and safe way for treating children having pulmonary hydatid cysts and living in the highly endemic areas with the least possible complications. This study included characteristics on presentation, investigation and operative techniques and postoperative morbidity and mortality in a group of patients with pulmonary hydatid cysts.

**Results:** fifteen patients with ages less than 6 years had cysts with a mean size of 14cm, ten patients with ages from 6-11 years had cysts with a mean size of 8.5cm and the last five patients with ages from 11-16 years had cysts with a mean size of 7.5 cm. Three patients had prolonged air leak for more than 2 weeks, and 3 patient developed empyema. No recurrence happened during one year follow-up. There were mortality.

**Conclusion:** the optimum approach for management of such children having pulmonary hydatid disease and living in highly endemic areas with limited facilities, especially the lack of postoperative intensive care unit is complete excision of the cysts whenever possible with maximum conservation of lung tissue.

**Index Word:** Pulmonary hydatid cysts, optimum management children.

### INTRODUCTION

Although most recorded cases of cystic *Echinococcus* are in adults, most of human infections occur during childhood and adolescence, the incidence of the disease and the need for surgical treatment is higher in rural populations than in urban

populations, particularly among children and adolescents<sup>1,2</sup>.

The cysts are found more frequently in the lungs of children and adolescents than their liver, while most

cysts found in adults are hepatic and relatively few are in the lungs<sup>1</sup>, this is explained by difference in the nature of tissue between the liver and the lung, and by longer resistance of lung tissue than the liver. This also explains the high incidence of giant pulmonary hydatid cysts and their multiplicity<sup>2</sup>. The cyst is identified on X-ray as a round or oval homogenous opacity that can be differentiated from pulmonary parenchyma<sup>3</sup>.

Diagnosis of pulmonary hydatid cysts is an indication for surgery which is the corner stone in the management, aiming at removing the cysts and preserving as much of tissue as possible<sup>2</sup>.

In the present study, stress was made on the special characteristics of pulmonary hydatid cysts in children living in highly endemic areas and the optimal surgical procedure applicable to them, in comparison with other series.

## PATIENTS AND METHODS

Thirty children with 40 pulmonary hydatid cysts were managed over a period of thirty six months from January 2007 to December 2009, their ages ranged from 24 months to 16 years with a mean age of 8 years, they were treated surgically at Al-Salam Hospital, Sada, North of Yemen and included 18 males and 12 females.

All patients were evaluated clinically and investigated by chest radiography (Fig. 1,2), chest computed tomography and abdominal ultrasonography (Fig.3). Serological studies were not available, and were not done for our cases. Diameters of the cysts were determined by radiological studies of the chest, and by operative findings.

Six cases with complicated (ruptured) cysts were managed preliminarily by (a) maintenance of patent tracheobronchial tree by oro-tracheal suctioning (b) Decompression of the pleural cavity for hydropneumothorax (c) Intravenous hydrocortisone to guard against anaphylaxis (d) Antibiotics given intravenously to guard against infection. Whenever the acute attack was over, surgical intervention was done in the same way as patients with uncomplicated cysts.

## Operative procedure:

All cases were performed under general anesthesia with a double lumen endotracheal tube. Intravenous hydrocortisone was given as prophylaxis against anaphylaxis. The patients were positioned in the lateral decubitus position and standard posterolateral thoracotomy was performed through the fifth intercostal space of the affected side. Pleural adhesions to the cysts were first released all around especially from chest wall, then packs soaked with hypertonic saline (H.S) 20% were placed all around to avoid contamination of the pleural cavity if a cyst ruptured (Fig. 4). An 18-Gauge needle connected to 30ml syringe was inserted in the upper most part of the cyst and the fluid was completely aspirated (Fig. 5). No scolical agent was injected into the cyst cavity. Insertion site was enlarged by cutting the pericystic layer with electrocautery, so that the germinative membrane was easily taken out and the bronchial openings encountered (Fig.6,7) these bronchial openings were closed with 2/0 silk sutures.

Different surgical techniques were applied to deal with the pericystic layer and the cyst cavity, depending on the extent of the pericystic layer, the cavity size and depth. Table 4.

(a) Cystotomy was done in 9 cases, in which the pericystic layer was minimally removed, the cavity was obliterated with imbrications sutures from within (capitonnage) using chromic catgut sutures (Fig.8).

(b) Partial pericystectomy, was done in 5 patients with interlobar and peripherally located cysts, in which excision of the great part of pericystic layer with small shallow cavity was done.

(c) Partial pericystectomy and capitonnage to obliterate the residual cavity was done for 10 patients

(d) Segmentectomy was done in 3 patients with giant cysts involving a large part of lung lobe.

(e) Lobectomy was performed for 2 patients with huge cysts completely involving a lung lobe, one case in upper left lobe, and the other case in right lower lobe.

Ten ml zylcaine 1% and 10ml marcain 0.25% infiltration were used routinely along the ribs above and below the thoracotomy space. One intercostal tube (24-30 F) with side holes was inserted, the tip of the tube was placed at the apex of the lung and the lower hole positioned at base of the lung. The tube was connected to an underwater seal and kept in situ for 3-12 days.

-Intravenous antibiotics: cefotaxin 50-100mg/kg/day divided into 2 doses and Amikacin 5mg/kg/dose were given 3 times daily, both were started 1 hour before operation and continued for 4-7 days.

Bilateral pulmonary hydatid cysts were found in 10 patients and each patient needed 2 thoracotomies, performed on 2 separate operations with a period 3 months interval.

Associated hepatic hydatid cysts with pulmonary cysts were found in 3 patients, one of them had left lung cysts and the second had right upper lobe cyst both of them needed thoracotomy followed by laparotomy few months later. The third patient had right lower lobe cyst and was managed in one session with right side transthoracic phrenotomy

## RESULTS

The patients were classified according to age into:

**Group (A):** Includes the youngest and largest number of patients (50%), most of them have giant hydatid cysts with mean cyst size 14cm.

**Group (B):** Includes (33.3%) of patients with cyst mean size 8.5cm.

**Group (C):** Includes oldest patients, representing (16.7%) of the cases with cyst mean size 7.3cm (table 1).

**Table (1): Age distribution of the patients and the size of the cysts**

	Age	No of patient	Cyst size	Mean
Group (A)	2-6 y	15	8-23cm	14cm
Group (B)	6-11 y	10	5-14cm	8.5cm
Group (C)	11-16 y	5	6-11cm	7.3cm

Most of the patients were males (62.5%) and 37.5% were females. Most of the patients were symptomatic with cough being the commonest symptom, hydatoptysis (expectoration of grape skin like materiel) occurred in 2 cases, allergic reaction were note observed in any patient. (Table 2)

**Table (2): Percentage frequency of symptoms:**

Symptoms	No. of patient	Percentage
Cough	9	30%
Cough + fever	5	16.6%
Hemoptysis	6	20%
Chest pain	3	10%
Asymptomatic	5	16.6%
Hydatoptysis	2	6.6%

Radiological examinations showed 40 cysts in 30 patients, 25 of which were on the right side (15 in lower lobe, 4 in middle lobe and 6 in the upper lobe). On the left side, there were 15 cysts (9 in lowers lobe, 6 in upper lobe), where bilateral cysts occurred in 2 patients. Ultrasonography of the abdomen showed associated Liver cysts as well.

**Table (3): Percentage frequency of radiological findings:**

Findings	No. of patients	Percentage
Found opacity	15	50%
Cavity with air fluid level	8	26.6%
Hydropneumo thorax	4	13.3%
Pleural effusion	3	10%

The management of pericystic layer and the cysts cavity were variable, according to the cyst site, depth of the cavity, extent of the pericyst layer and how much it involves the lung lobe: cystotomy and capitonnage was applied in 9 cases, partial pericystectomy in 5 cases, partial pecystectomy and capitonnage in 10 cases, segmentectomy in 3 case, lobectomy in 3 cases (table 4).

**Table (4): Operative procedure:**

A) Cystotomy + capitonnage	9	30%
B) Partial pericystectomy	5	16.6%
C) Partial pericytectomy + capitonnage	10	33.3%
D) Segmentectomy	3	10%
E) Lobectomy	3	10%



Fig. 1: Plain X-ray chest shows huge hydatid cyst left lung.



Fig. 2: Plain X-ray chest shows huge hydatid cyst in the right lung



Fig. 3: C.T. chest shows huge hydatid cyst in the left lung



Fig. 4: Hydatid cyst in the lung



Fig. 5: Aspiration from a hydatid cyst

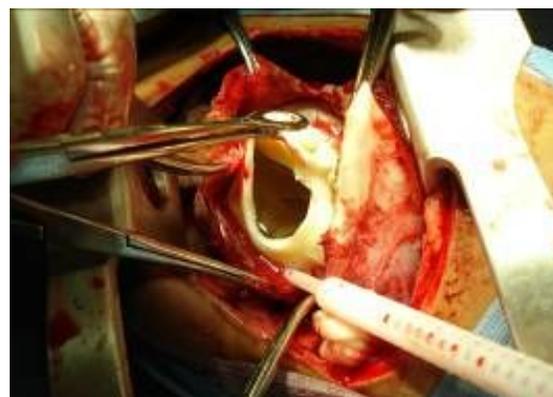


Fig. 6: Removal of a hydatid cyst



Fig. 7: After removal of a hydatid cyst



Fig. 8: Pericystectomy with capitonnage



Fig. 9: Plain X-ray shows removal of a hydatid cyst from the left lung with chest tube



Fig.(10: Plain X-ray shows removal of a hydatid cyst from the right lung with chest tube

### Mortality and morbidity:

There were no mortality. Postoperative prolonged air leak for more than one week occurred in 3 patients (10%), two of them improved with keeping the chest tube, and chest physiotherapy, the third one was reoperated, because of increasing air leak due to bronchopleural fistula and had smooth postoperative recovery. Another patient developed postoperative pleural effusion and a chest tube was reinserted with improvement of the effusion. Follow-up was carried out for all cases with repeated clinical and radiological assessment over of one year and there was no recurrence on the same side of the operation, only one patient got another pulmonary hydatid cyst, 6 months postoperatively on the opposite chest side and contralateral thoracotomy was done for him.

## DISCUSSION

Hydatid disease is still a national problem in highly

endemic countries like Yemen and needs epidemiologic prevention for its eradication. Surgery is the best choice in management, the role of medical therapy is not well established<sup>13</sup>, so, we did not rely on the medical in the management of our cases.

The predominance in boys (62.5%) over girls (37.5%) among this study is mostly due to behavioral difference between both sexes, with more exposure of boys. This is confirmed in other series<sup>14,15,16</sup>. However, in Bulents report incidence of hydatid cysts was equal in males and females<sup>7,8</sup>.

Lung involvements tends to decline with age<sup>1</sup>, this corresponds with this study as patients of group (A) represent 50% of the cases, group (B) represent 33.3% of patients, group (C) represent 16.7% of the cases. Table 1.

The age of the youngest patient in this study was 21 months, while in Elburjo study<sup>(17)</sup>, it was three years and in Halezeroglu study<sup>13</sup> it was 9 years. In Ali et al study<sup>(3)</sup>, it was 2 years old and Prashant has reported

hydatid in 6 months old infant <sup>9</sup>.

In this study, the finding that there is higher incidence of pulmonary than hepatic hydatid cysts in children and adolescents corresponds with other studies <sup>3,5</sup>. The explanation for that is the liver is a compact organ and the hepatobiliary capsules limits the cysts growth and the low resistance of lung tissue provides an excellent medium for rapid growth of hydatid cysts. However, Sehitoqullari<sup>(4)</sup> believed that liver cysts were more common in childhood and Talaiezadeh <sup>6</sup> found nearly equal incidence of hydatid cysts in lung & liver in children.

Burgos <sup>18</sup> defined pulmonary hydatid cysts as giant if it equals nearly half of hemithorax. It is clear that giant cysts were more common young age group, table (1), this has been explained by the more elastic properties of lung tissue in group children, this explanation similar to other studies such as Ali. et al <sup>3</sup>.

Cough was the most common symptoms in our patients similar to other studies <sup>5,13,14</sup>. But in Talaiezadeh <sup>6</sup> series, chest pain was the most common symptom.

Diagnosis of pulmonary hydatid cyst in endemic area should be suspected in any patient presented with single or multiple opacities on the chest X-ray <sup>19</sup>. The radiological findings in this study are similar to that in Elburjo of study<sup>17</sup>.

Conventional X-ray and computed topography image were applied to all cases and there was 100% accuracy in the diagnosis of all cases, however, the accuracy of radiology was 99% in Saleh et al <sup>21</sup>, 94.44% in Ali et al <sup>3</sup>, 84% in Montazeri series <sup>5</sup>, and 96.4% in koeseoglu series <sup>23</sup>.

The multiplicity of cysts was higher in this study (40 cysts in 30 cases than Halezeroglu <sup>13</sup> series (50 cysts in 47 cases). Bilateral pulmonary hydatid cysts were found in 4 cases (13.3%) of this study, where was no bilaterality in Halezeroglu <sup>13</sup> series.

Criteria for selecting operative technique differ from one country of another and are closely related to experience with disease and the condition of the cyst <sup>22</sup>. In this study, much concern was directed to preserve pulmonary tissues on dealing surgically with these cases because:

- (1) Children have long life expectancy.
- (2) Possibility of re-infection is high especially with lack of prevention programs.
- (3) Most of these children have huge cysts and

resection may lead to postoperative respiratory complications.

(4) Bilateral cysts are not uncommon in the endemic areas.

(5) Most of these patients are not that healthy and major lung resection may lead to considerable hazards.

(6) Lack of postoperative intensive care and endobrochial endoscopy for care of patients after major resection, so resection was avoid in this study unless the lobe was thoroughly destroyed; accordingly, we performed lobectomy in only 3 cases and segmentectomy in 3 cases.

Parenchyma saving procedure was applied in 24 patients of this study (80%).

Safety rules were strictly followed (1) selective intubations of the opposite lung (2) packs soaked in H.S 20% were applied all around the cysts.

This controlled evacuation avoids contamination possibility of rupture and lower the intra-cystic pressure leading to easier and safer mobilization of the pericyst layer (Fig. 2). After that, the choice of the procedure depended on local factors: (1) cystostomy and capitonnage, applied in 9 cases with cysts having small pericyst layer with deep residual cavity; (2) partial pericystectomy applied in 5 cases with cysts having large pericyst layer and superficial residual cavity, they did not need capitonnage as cysts were interlobar and peripherally located with small residual <sup>3</sup>; partial pericystectomy and capitonnage was applied in 10 cases partial pericysted <sup>20,21</sup> resulted in big residual cavities which were obliterated with capitonnage.

Patients presented with complications were treated first from the acute attack followed by thoracotomy with conservative resection, which is similar to other series <sup>19</sup>.

Cases with bilateral pulmonary hydatid cysts were managed with staged resection, same as in Shamji series <sup>19</sup>. The lack of ICU made the staged resection with 2 thoracotomies 3 months apart the procedure of choice for our patients with good outcome. In other series, the median sternotomy approach was preferred <sup>21</sup>

In this study, one stage surgical management of lung and liver hydated cyst by right thoracotomy and phrenotomy was done. This approach was performed by Biswas et al <sup>24</sup>, and Ali et al <sup>3</sup>.

There was no mortality in this study, same as Elburjo series<sup>17</sup> and Halezerogh<sup>13</sup>. postoperative complications occurred in 3 cases (13.3%) of this study, three of them improved with conservative treatment, only one needed re-operation for closure of bronchopleural fistula with good final outcome. There was not any recurrence.

## CONCLUSION

Complete excision of pulmonary hydatid cyst with maximum preservation of lung tissue is an appropriate approach for children with pulmonary hydatid cyst in highly endemic areas with high incidence of giant cysts, bilateral lung involvement, especially if I.C.U is not available .

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