# **Comparison of Pulmonary Hydatid Disease between Children and Adults**

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#### ABSTRACT

**Background:** Hydatid disease is a parasitic infestation which is endemic in many sheep and cattle raising areas and is still an important health hazard in the world. The aim of this study was to compare the clinical features, radiologic accuracy, and the surgical approaches of pulmonary hydatid cysts among children and adults in Northwestern Iran.

**Materials and Methods:** 445 patients treated for pulmonary hydatid cysts over the last ten years, were retrospectively evaluated. The patients were arranged in two groups. There were 383 adults (86%) and 62 children (14%) and their clinical, radiological and surgical characteristics of the pulmonary hydatid cysts were compared.

**Results:** Unlike adults, in children the frequency of pulmonary hydatid cysts in males was significantly (p=0.0036) higher than in females (65% vs. 36%). Cough was the most common symptom in both groups. The frequency of hemoptysis was more common (p=0.0042) in adults (38%) than in children (19%). However, giant pulmonary hydatid cysts were more commonly found in children (p<0.05). Cystotomy with or without capitonnage was the most preferred method used in both groups. Postoperative complications (pneumothorax) developed in five adults and one child, and were more frequent in non-capitonnage procedures. Only one recurrence was observed in the adult group. The accuracy of chest x-ray was 84% and 74% in children and adults, respectively.

**Conclusion:** Due to the high accuracy of chest x-ray in diagnosis of hydatid cysts of the lung in both groups, it is recommended as the preferred method of diagnosis in endemic regions. **(Tanaffos 2007; 6(1): 13-18) Key words:** Hydatid cyst, Lung, Chest x-ray, Adult, Children

INTRODUCTION

Hydatid disease is an important public health problem in Iran (1). It is common in rural areas where cattle and dogs are kept. The primary hosts of the *Echinococcus granulosus* tapeworms were dogs and canines. They produce eggs in the intestine and pass them in the stool. Eggs are ingested by

Address: Department of Thoracic Surgery, Imam Khomeini Hospital, Tabriz Medical University, Tabriz-Iran Email address: sokouti\_m@yahoo.com intermediate hosts such as sheep and cows. Humans become accidental hosts by eating tapeworm eggs (2). In children, the lungs are the most common organ infected by larval form of *Echinococcus granulosus* (64% in children compared with 20-30% in adults). Cysts may grow faster in the lungs than in the liver due to less elasticity of the lungs. This may explain the high incidence of disease in these organs in children (3, 4). In the present retrospective comparative study, the clinical features, surgical approaches and radiologic accuracy of pulmonary

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hydatid cysts in children and adults have been investigated.

## **MATERIALS AND METHODS**

This study was conducted on hospitalized patients who were operated for pulmonary hydatid cysts at the Imam Khomeini Medical Center in Tabriz, Iran during 1995 to 2005. Of 445 patients, 62 patients (14%) were in the pediatric age group (<12 years old with a men age of 10.5 years ranging from 3 to 12 vrs.) and 383 patients (86%) were adults (>20 years old with a mean age of 46 years ranging from 20 to 76 yrs.). Data were collected from the medical records of the patients in the archives and analyzed in terms of age, radiologic findings, symptoms and sings of presentation. At presentation, chest radiography was performed in 94% of the patients. Casoni's intradermal tests, serologic antibody tests of hydatid cysts, and eosinophil counts were not used routinely because of their low diagnostic value compared with radiologic techniques.

Patients who had complicated or uncomplicated hydatid cysts of the lung were enrolled in this study. All patients were fit for general anesthesia and the pulmonary hydatid cysts were diagnosed or suggested by means of chest x-ray. The patients with cardiopulmonary disorders and hemoptysis for other reasons were excluded from the study.

Cystotomy and partial pericystectomy with or without capitonnage were used as the preferred surgical methods for unruptured hydatid cysts. All cysts were delivered except ruptured cysts. An incision was made in the pericyst, and the cysts were delivered intact. Parts of the pericyst protruded outside the lung parenchyma were excised. Four giant cysts were aspirated by needle and then evacuated. Bronchial communications were closed with fine resorbable sutures. In four cases (three adults and one child) in which the whole lobe of the lung had been destroyed, lobectomy was performed. There were eleven cases of empyema (7 adults and 4 children) in which decortication was carried out. Then, all patients were followed up in surgical outpatient clinic by the surgeon who had operated the patient and all the relevant information was recorded in their files for at least two years. The comparison between the groups was made using the binomial proportion test. The study was approved by the medical ethics committee of Tabriz University of Medical Sciences.

# RESULTS

Out of 62 children, 40 (64%) were males and the remaining 22(36%) were females. The corresponding values in the adult group were 207 (54%) and 176(46%), respectively. Statistical analysis indicated that in children, males are more likely to be infected with this disease (p=0.0036). Although a similar manner was observed in adults, it was not statistically significant (p=0.242). The most common symptom was cough followed by mucopurulent sputum. The frequency of hemoptysis was 38% in adults and 19% in children indicating that this symptom is more common in adults than in children (p=0.0042). Other symptoms observed were chest pain, febrile episodes and respiratory distress; however, no differences were found between adults and children with respect to these symptoms (Table 1).

 Table 1. Symptoms of pulmonary hydatid cyst disease in adults and children (n=445).

	Adults (n=383)	Children (n=62)	p-value
Cough	302 (79%)	58(93%)	0.0063
Hemoptysis	146(38%)	12(19%)	0.0042
Chest pain	225 (59%)	42(68%)	0.18
Febrile episodes	183 (48%)	23(38%)	0.118
Mucopurulent sputum	259 (68%)	35(56%)	0.085
Respiratory distress	165 (43%)	35 (56%)	0.05

Ninety-three percent of patients were examined by chest radiography. Diagnostic accuracy of chest radiography was 84% (p<0.05) in children and 74% in adults (p<0.05). Radiologic appearances of pulmonary hydatid cyst in both groups are shown in table 2 and figures 1-5. **Table 2.** Radiographic appearances of pulmonary hydatid cyst in adults and children (n=473 cysts)

	Adults (n=403)	Children (n=70)	p-value
Notch sign	172 (43%)	34 (49%)	0.359
Water lily (Camellote)	37(9%)	5(7%)	0.58
Meniscus (moon)	4(1%)	5 (7%)	0.0005
Migratory sign	6(2%)	2(3%)	0.413
Combo (double air)	0(0%)	3 (4%)	0.00003
Annular solar eclipse	6(2%)	2(3%)	0.651
Pneumothorax	13( 4%)	3(4%)	0.651

Table 3. Location of pulmonary hydatid cysts in adults and children (n=473 cysts)  $% \left( n=473,n=1\right) \left( n=1\right) \left$ 

	RLL	LLL	RML	RUL	LUL	Lingula
Children	29	24	7	5	4	2
(n=70)	(42%)	(34%)	(10%)	(7%)	(5%)	(3%)
Adults	139	110	44	37	23	12
(n=403)	(35%)	(27%)	(11%)	(9%)	(8%)	(3%)
p-value	0.263	0.231	0.819	0.58	0.998	0.959

RLL= Right Lower Lobe RML= Right Middle Lobe

LUL= Left Upper Lobe

RUL= Right Upper Lobe LLL= Left Lower Lobe Lingula= Lingular Lobe



Figure 1. Notch Sign in chest-x-ray



Figure 2. Combo sign (Double air layer appearances)

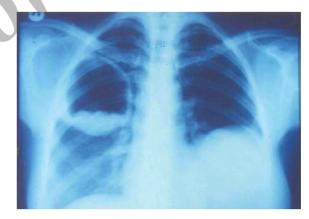


Figure 3. Water Lily sign (Camellote sign)

In adults, 55% of cysts were observed in the right lung. Almost the same value (58%) was found in the children group (Table 3). A difference was observed in the occurrence of intact hydatid cysts in children (n=40, 65%) and adults (n=134, 35%) which was statistically significant (p<0.05). Giant cysts (greater than 15 cm) were more prevalent in children (29%) than in adults (3%) (p<0.05). One of these giant cysts is shown in figure 6.

Synchronous pulmonary and liver involvements were observed in 16% of children and 28% of adults which was not statistically (p=0.0501) different.

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Also, no significant difference was detected regarding the characteristics of the hydatid cysts in both groups (Table 4).



Figure 4. Moon sign in chest-x-ray



Figure 5. Aunular solar eclipse sign in chest-x-ray.



Figure 6. Macroscopic appearance of intact giant hydatid cyst of the lung.

 Table 4. Comparison of the characteristics of the hydatid cysts in both

 groups (n=445)

Single		One- sided	Bilateral	
	cysts	multifaceted cysts	cysts	
Adults (n=383)	256 (67%)	88 (23%)	39 (10%)	
Children (n=62)	40 (65%)	15 (24%)	7 (11%)	
P-value	0.719	0.657	0.790	

Seven adults and four children presented with pneumothorax. After insertion of thoracic tube, drainage and improvement of respiratory distress, pulmonary hydatid cyst was inspected. In four adults and three children, there was a need for decortication because of pleural thickening.

All patients were managed by surgery. A standard thoracotomy and rarely anterior thoracotomy, cystotomy and delivery of the cysts, partial pericystectomy with (4%) or without capitonnage of the residual pericyst cavity were performed on the affected side. Segmental resections of the lung and lobectomy were carried out in six and four patients, respectively. Related hepatic cysts were not operated on at the same time. They were treated at least one month after thoracic surgery. Four patients with gaint cysts were operated via evacuation, cystotomy and partial pericystectomy of the residual pericyst cavity. Of 445 subjects, only in six patients (1%) were postoperative complications observed. Delayed pneumothorax was seen in five patients after operation (four adults and one child). Anaphylactic shock occurred only in one adult patient. The patients were followed up for 2-5 years and no mortality was observed among the operated patients. There was only one recurrence of intact hydatid cyst; however, among the cases with ruptured hydatid cyst, six recurrences were seen.

## DISCUSSION

Hydatid disease in children and adults is a major health hazard in Northwestern Iran and is considered an endemic disease in this area (1). Consistent with other reports (2, 4), the results indicated that the pulmonary hydatid cyst is more common in children and young males than in females, although this was found to be significant only in the former group. In our study, 62 of 445 patients diagnosed with pulmonary hydatid cyst were under the age of 12 (14%). The compressible nature of the lung tissue allows the cyst to reach a large size before symptoms appear in children (4, 5). Most symptomatic cysts are larger than 5 cm in diameter (2). Hepatic hydatid cyst is common in older patients, whereas the lungs are commonly affected in childhood (3, 6). The most common symptoms of pulmonary hydatid disease are cough, mucopurulent sputum, and chest pain (5). In our study, although cough and chest pain were the most common frequent symptoms in children, hemoptysis was encountered less (19%). Due to the 65% incidence rate of intact hydatid cyst in children and 35% in adults shown in our study, rupture of pulmonary hydatid cyst occurred less in children. The ratio of unruptured cysts is 3:1 in children and the opposite is true in adults (7).

The diagnosis of pulmonary hydatid cyst by radiographic appearances of chest x-ray can be more easily made in children than in adults (7-10).

Serologic methods have false negative results (10% in adults and 30% in children) and low accuracy (40%) in pulmonary echinococcosis (7). Therefore, these tests were not routinely used in our study. The frequency of the radiographic appearances of pulmonary hydatid cyst of children and adults are shown in table 2. In addition, in figure 1, the radiographic appearances of these cysts have been illustrated. The radiologic signs were diagnosed as follows: notch sign, figure 1 (11); combo sign or double air layer appearances, figure 2, that both are specific for pulmonary hydatid disease (11,12); water lily sign or Camellote sign, figure 3 (14) that are pathogenomonic for pulmonary hydatid cyst; moon, figure 4, and annular solar eclipse signs, figure 5 (13,15). Although ultrasound of the thorax and abdomen is useful for diagnosis of pulmonary hydatidosis (9). As in 50% of the cases the ultrasound was not used, the relevant data was not considered in the present study.

Treatment of hydatid cyst is surgical and children usually tolerate it well (16). The use of albendazol is restricted in children under the age two (17). The resection rate in lung cysts has been reported as 6-21% in different series (4, 5, 18). In this study, wedge resection was performed for small cysts found at the edges of the lobe. In case of wedge destruction, segmentectomy was carried out. When lung cysts were found centrally or there was destruction of the lobe, lobectomy was used. The resection rate in our patients was 2%.

Recurrence has been reported in the literature to be 7-14% and is due to the ruptured hydatid cyst and the type of operation (13, 19, 20). Our recurrence rate was 1%. This low rate of recurrence observed in this study could be the result of delivery of the cyst or the surgical methods.

## CONCLUSION

According to the present study, chest x-ray can be

recommended as a diagnostic tool for hydatid cysts in endemic areas due to its high accuracy.

### REFERENCES

- Harandi MF, Hobbs RP, Adams PJ, Mobedi I, Morgan-Ryan UM, Thompson RC. Molecular and morphological characterization of Echinococcus granulosus of human and animal origin in Iran. *Parasitology* 2002; 125 (Pt 4): 367-73.
- King CH. Cestodes (tapeworms): Mandell GL, Bennet JE, Dolin R. Principles and practice of infectious disease 6<sup>th</sup> ed. New york: Churchill Livingston, 2005; 3290-2.
- Beggs I. The radiology of hydatid disease. AJR Am J Roentgenol 1985; 145 (3): 639-48.
- Karaoglanoglu N, Kurkcuoglu IC, Gorguner M, Eroglu A, Turkyilmaz A. Giant hydatid lung cysts. *Eur J Cardiothorac Surg* 2001; 19 (6): 914-7.
- Kilani T, El Hammami S. Pulmonary hydatid and other lung parasitic infections. *Curr Opin Pulm Med* 2002; 8 (3): 218-23.
- Symbas PN, Aletras H, Harlafitis NN. Hydatid disease of the lung. In: Shields TW, et al. General Thoracic Surgery, 6<sup>th</sup> ed. Philadelphia: Lippincott Wilkins, 2005; 1298-1307.
- Tuazon AO, Paster comp H. Pulmonary hydatidosis in Chernek V, et al. Kendig's disorders of the respiratory treact in children. 5<sup>th</sup> ed. W B. Saunders Company, 1990; 867-72.
- Cangir AK, Sahin E, Enon S, Kavukeu S, Akay H, Okten I, et al. Surgical treatment of pulmonary hydatid cysts in children. *J Pediatr Surg* 2001; 36 (6): 917-20.
- Hafsa C, Belguith M, Golli M, Rachdi H, Kriaa S, Elamri A, et al. Imaging of pulmonary hydatid cyst in children. J Radiol 2005; 86 (4): 405-10.
- Dogan R, Yuksel M, Cetin G, Suzer K, Alp M, Kaya S, et al. Surgical treatment of hydatid cysts of the lung: report on 1055 patients. *Thorax* 1989; 44 (3): 192-9.
- McPhail JL, Arora TS. Intrathoracic hydatid disease. *Dis Chest* 1967; 52 (6): 772- 81.
- Bloomfield JA. Protean radiological manifestations of hydatid infestation. *Australas Radiol* 1966; 10 (4): 330-43.

- Jerray M, Benzarti M, Garrouche A, Klabi N, Hayouni A. Hydatid disease of the lungs. Study of 386 cases. *Am Rev Respir Dis* 1992; 146 (1): 185-9.
- Kennedy D, Sharma OP. An unusual presentation of hydatid disease of the lungs. *Chest* 1990; 97 (4): 997-9.
- Burgos L, Baquerizo A, Munoz W, de Aretxabala X, Solar C, Fonseca L. Experience in the surgical treatment of 331 patients with pulmonary hydatidosis. *J Thorac Cardiovasc Surg* 1991; 102 (3): 427- 30.
- Elburjo M, Gani EA. Surgical management of pulmonary hydatid cysts in children. *Thorax* 1995; 50 (4): 396-8.
- Laurance L. Bruton JL. Keith L. Goodman& Gilman's. The Pharmacological Basis of Therapeutics. 11<sup>th</sup> ed. Mc Graw Hill Co, 2006; 1082.
- Aytac A, Yurdakul Y, Ikizler C, Olga R, Saylam A.
   Pulmonary hydatid disease: report of 100 patients. *Ann Thorac Surg* 1977; 23 (2): 145-51.
- 19. Xu MQ. Hydatid disease of the lung. *Am J Surg* 1985; 150 (5): 568-73.
- Perianayagam WJ, Freitas E, Sharma SS, Mulalidharan S, Jairaj PS, John S. Pulmonary hydatid cyst: a 25-year experience. *Aust N Z J Surg* 1979; 49 (4): 450-3.