

## Characteristics of pediatric pulmonary hydatid cyst in Mashhad, North East of Iran.

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### ABSTRACT

Hydatid disease is an important health problem in endemic areas like Iran. Unlike adults, the occurrence of pulmonary hydatid cyst is greater than hepatic cysts in pediatric population.

In this study we evaluated the characteristics of pediatric pulmonary hydatid cyst in our region. We reviewed the medical records of all children with the diagnosis of pulmonary hydatid cyst in Dr Sheikh's children hospital between 2015-2019. 30 patients were enrolled during the study period. Diagnosis was confirmed using imaging and serology tests.

Mean age of patients was  $8.5 \pm 2.1$  years (range:6-14 years). 73% were male and 80% were living in rural areas. All patients were symptomatic at presentation and the most frequent symptom was cough (76%) followed by dyspnea (43%). The most frequent site of involvement was the lower lobe of the right lung (30% of patients) and bilateral involvement was seen in 16.7% of patients. 13.3% of patients had simultaneous hepatic involvement. Complications were reported as follows: pleural effusion 30%, superinfection and abscess formation 16.7% and pneumothorax in one patient. All patients underwent surgical treatment. Lobectomy was performed in 5 cases (16.6%). Pulmonary hydatid cyst must be considered in children with respiratory symptoms in endemic areas. Early diagnosis might lower the risk of complications and lobectomies.

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## Introduction

Hydatidosis is a common health problem in developing countries because of hygienic conditions (1). This disease is endemic in Iran and many cases are reported annually (2,3). It is a parasitic infection caused by *Echinococcus granulosus*, a tapeworm that lives in the intestines of canids in its adult form and infects humans as intermediate hosts and causes cysts (4).

Unlike adults, liver involvement is less frequent in children and the most frequently affected organs

by hydatid cysts are lungs (5,6). Children may be asymptomatic for a period of time after the initial infection but with the formation of cyst and its growth symptoms will ensue. Clinical features will depend on the location and size of the cyst. Very small cysts that are located peripherally may be asymptomatic and may be found incidentally on chest x rays but larger cysts usually cause symptoms due to the mechanical effects on the adjacent tissue or other complications (7,8).

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Plain chest x ray plays an important role in the diagnosis of pulmonary hydatid cyst (9,10). Chest CT scan and MRI are also useful diagnostic modalities for the evaluation and diagnosis of pulmonary hydatidosis (11,12).

Ultrasound has also proven useful in the diagnosis of pulmonary hydatid cysts especially for the peripherally located cysts. It is an easy, non-invasive method for diagnosis confirmation (13).

The mainstay of therapy in pediatric pulmonary hydatid cyst is surgery but medical therapy might also be used in selected patients with small cysts or contraindications to surgery (14,15).

This study aims to evaluate the characteristics of pediatric pulmonary hydatid cyst in Mashhad, North East of Iran.

## Methods

This was a retrospective chart review. All children under 16 years old with a diagnosis of pulmonary hydatid cyst who were admitted to Dr Sheikh's children hospital between March 2015 to May 2019 were included in the study.

This hospital is affiliated with Mashhad University of Medical Sciences and is the referral center for pediatric pulmonology and surgery in East of Iran.

## Results

30 patients with a diagnosis of pulmonary hydatid cyst were hospitalized in Dr Sheikh's children hospital during the study period. Demographic characteristics are shown in table 1.

**Table 1 :** patient's characteristics.

<b>Sex</b>	male	N=22 (73%)
	female	N= 8 (26%)
<b>Area of residence</b>	city	N=6 (20%)
	rural	N= 24 (80%)
<b>Age</b>	8.5±2.1 years	
<b>weight</b>	31.2±12.3 kg	
<b>Height</b>	134±14.2 cm	

All 30 patients were symptomatic at the time of diagnosis. Cough and dyspnea were the most common patient's complaints. Table 2 summarizes the frequency of patients' symptoms at the time of diagnosis. 4 patients had concomitant involvement of lung and liver.

Postero-anterior chest X ray was performed for all patients and it showed lung involvement as follows: the most frequent involvement was in the lower zone of the right lung (9 cases or 30%).

Bilateral lung involvement was seen in 5 patients (16.7%). Ultrasonography or chest computed tomography was used for the confirmation of diagnosis in combination with serology testing (IHA).

**Table 2.** clinical symptoms of patients at the time of admission.

primary symptom	Number of patients	percent
Cough	23	76.7
Dyspnea	13	43.3
Fever	11	36.7
Hemoptysis	6	20
Chest pain	4	13.3
Abdominal pain	3	10
Purulent sputum	3	10
Nausea and vomiting	1	3.3
urticaria	1	3.3
Weight Loss	1	3.3

Ultrasonography was performed for all patients and confirmed the diagnosis of hydatid cyst in 17 patients. 2 of them reported as perforated hydatid cyst. 9 patients had pleural effusion.

5 patients sonogram was reported as having pulmonary abscess. All patients underwent CT examination of the chest for better localization of the cyst and also detecting other cysts that might not have been shown in chest x ray or sonograms.

Results of the chest CT scans are shown in table 3.

**Table 3.** Frequency distribution of involvement of different lobes of the lung in patients' chest CT scans

Involved lobes of lunged in CT	Number of patients	Frequency percentage
inferior lobe of the right lung	8	26.7
superior lobe of the left lung	5	16.7
superior lobe of the right lung	4	13.3
inferior lobe of the left lung	4	13.3
middle lobe of the right lung	3	10
Bilateral inferior lobes	3	10
Bilateral superior lobes	3	10

Serology test of hydatid disease was checked for all patients using indirect hemagglutination test (IHA) and positive results was shown in 43.3% of cases. 4 patients who had concomitant involvement of lung and liver, all had positive IHA test. All patients underwent surgical treatment and after surgery, they received albendazole for at least 1 month (range 1 to 3 months). 23 patients received complete resection of the cyst. In 2 patients, the cyst was perforated and the perforated cyst was resected. 5 patients underwent lobectomy.

Post-operative complications occurred in 6 patients; Pneumothorax and collapse were each reported in 2 patients and abscess and fistula formation was reported in 2 patients. Mortality was not reported in any of these 30 cases.

## Discussion

Hydatid cyst is a zoonotic infection caused by the larva of *Echinococcus granulosus*. Humans are accidental intermediate hosts of this parasite. They ingest the eggs accidentally and then the eggs hatch into metacestodes which infest the internal organs most predominantly liver and the lungs (16).

Iran is one of the endemic areas for this infection and many cases of this infection are reported annually (17). Children also acquire this infection and the most prevalent affected organ in children is lung (5,6). No gender prevalence is recognized for this disease. But some studies have suggested that the infection is more prevalent in boys and this has been attributed to their being more in contact with animals (18,19).

In our study the prevalence of pulmonary hydatid cyst was greater in boys than girls too. Children living in rural areas are at increased risk of being infected due to their close contact with sheep and dogs as well as the unavailability of clean water supplies (20). In our study 80% of the affected children were residents of rural areas.

Pulmonary hydatid cyst can present with a variety of clinical manifestations. Cough, fever, dyspnea, chest pain and hemoptysis, have all been reported in patients with pulmonary hydatidosis (21,22).

In our study cough and dyspnea were the most prevalent symptoms and all patients were symptomatic at presentation. Simple chest x rays are usually the first imaging modality for the diagnosis of pulmonary hydatid cyst and they can give us information about the location of the cyst, whether it is ruptured or intact and presence of complications (23). In this study all patients had abnormal chest x rays. The most common site of involvement on chest x ray was opacity in the lower zone of the right lung.

Computed tomography scan (CT scan) was also performed for all patients for the confirmation of the diagnosis and better localization of the cysts.

X rays and CT scan are the two most used imaging modalities (23,24) and ultrasonography has been shown effective in the diagnosis of pulmonary hydatid cyst especially in the peripherally located cysts and is also useful for the evaluation of pleural involvement (13,23).

Wall sign has been described for the pulmonary hydatid cyst in sonography and is said to be nearly 100% specific for the diagnosis of pulmonary hydatid cyst (13). In our study chest sonography was performed for all patients and confirmed the diagnosis in 17 patients.

There is no highly sensitive and specific serological test for the diagnosis of pulmonary hydatid cyst (25). Indirect hemagglutination test (IHA) and ELISA test are the two frequently used tests for the detection of cystic echinococcosis. Their sensitivity in

the detection of pulmonary disease is lower than hepatic involvement and false negative results are not uncommon (26,27).

Serological tests must be used in conjunction with imaging for the diagnosis of hydatid disease but they are often used for the monitoring of treatment (25). In this study, we had a positive IHA test in 43.3% of patients. It is important to notice that the test was positive in all patients with the simultaneous involvement of liver and lung.

The most frequent site of involvement in pulmonary hydatid cyst is usually the inferior lobes and right basal lobe is the most common (23,28). In our study, the inferior lobe of the right lung was the most frequently involved area (26.7%).

Surgery is the treatment modality of choice in pulmonary hydatid cyst (29,30), pharmacotherapy might be used in selected patients with small peripherally located cysts.

When surgical treatment is performed, it is important to try to save the lung parenchyma (30). In cases with more than 50% damage to the lung parenchyma or complications like abscess, hemorrhage or bronchiectasis lobectomy is performed (31). In our study all patients underwent surgical treatment. 25 patients received complete resection of the cyst and for 5 patients lobectomy was performed due to the large involvement of the lung or complications.

Complications after pulmonary hydatid cyst surgery is reported to be less than 1% in the literature and the most common complication is prolonged airleak (21,32). In our study, we had 20% postoperative complications. No mortality was observed in the present study which is compatible with the reports in the literature which have reported the mortality of hydatid cyst between 0-2% (30, 33).

## Conclusion

Pulmonary hydatid cyst must be considered in the differential diagnosis of children with pulmonary symptoms in endemic areas like Iran. Diagnosis can be made using imaging techniques in conjunction with serological testing.

Surgery remains the mainstay of therapy in pulmonary hydatid disease and mortality rates are very low. In endemic areas attempts should be made to prevent the disease.

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