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Massive Life-Threatening Hemoptysis from Pulmonary Hydatid Cysts: A 13 - Year Experience from an Endemic Area

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ABSTRACT

Background: *Echinococcosis is a parasitic disease often seen in sheep and cattle raised in countries with unsanitary conditions. The disease is encountered endemically in northwest of Iran. Life-threatening hemoptysis due to a pulmonary hydatid cyst is a rare condition. This study assessed the prevalence and treatment of this condition.*

Materials and Methods: *In a retrospective study, between 1993 and 2005, 520 patients with pulmonary hydatid cysts were operated at the Imam Khomeini Hospital in Tabriz, Iran. Ten (1.9%) cysts exhibited life-threatening hemoptysis (Group II). The remaining 510 patients with pulmonary hydatid cyst were studied (Group I) and compared with group II.*

Results: *Ten patients were included in group II, with a mean follow – up of 2.5 years. Four (40%) cases were males and six (60%) were females. The age ranged between 8 and 46 (mean 27.90 ± 13.86) years. The most common symptoms were cough (100%), massive hemoptysis (100%), respiratory distress (100%), and chest pain (90%). The amount of hemoptysis was between 600 and 1400 ml (mean 840 ± 249.66) a day. The origin of bleeding in six (60%) was from the cyst in the right lung and in four (40%) from the left lung. Bronchoscopy revealed the site of bleeding in all 10 patients, and the lobe in seven (70%). Chest x-ray and CT scan showed patchy infiltration in one or both lungs and a ruptured hydatid cyst in eight cases. Lobectomy was the most frequent applied procedure (100%). Two (20%) cases died due to blood aspiration and respiratory distress syndrome, one in the 3rd and the other in the 4th postoperative day. The postoperative hospital stay ranged between 7 and 25 (mean 11.30 ± 6.56) days. Two and a half years follow-up of patients was good. There was no recurrence. There were significant differences between the two groups In terms of complications and mortality ($\chi 2$ =102.39, df=1, p<0.0005) and ($\chi 2$ = 5.82, df=1, p<0.16).*

Conclusion: *Massive life-threatening hemoptysis due to pulmonary hydatid cysts must be managed as an urgent clinical entity. It has serious symptoms, serious post-operative complications and a high mortality rate.* ***(Tanaffos 2008; 7(3): 41-46)***

Key words: *Massive Hemoptysis, Pulmonary hydatid cyst, Echinococcosis*

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INTRODUCTION

Hydatid disease is a parasitic tapeworm infection of the genus Echinococcus**,** which is still endemic in many sheep and cattle raising areas**.** It is still an important health problem throughout the world (1). Infestation is confined to geographical areas like Australia, New Zealand, South Africa, and the Mediterranean region, Europe, Asia and Africa (where there is continuous contact between humans and certain domestic carnivores such as dogs and sheep) (2, 3). The lung is the second most common site for hydatid disease after the liver. When hydatid cysts lie peripherally, they usually remain asymptomatic and may be unrecognized for a long time. Massive hemoptysis is life threatening and is one of the rare complications and fatal manifestations of pulmonary hydatid disease. Surgery is the primary treatment for patients with pulmonary hydatid disease (3, 4). Life-threatening hemoptysis due to pulmonary hydatid cysts often results in death before arriving to a medical facility. A high morbidity and mortality rate follows patients with this condition. Massive hemoptysis due to this disease is rare. Its incidence is between 0%- 0.9% in the literature (3, 5). This study focused on the importance of urgent surgery and management of massive hemoptysis due to pulmonary hydatidosis in an endemic area.

MATERIALS AND METHODS

This retrospective study assessed hospitalized patients operated for life-threatening hemoptysis due to pulmonary hydatid disease at the Imam Khomeini Medical center in Tabriz, Iran from 1993 to 2005. Of 520 patients, ten patients (4 men and 6 women) in group II with a mean age of 27.90 ± 13.86 years (range 8 to 46 years) had life-threatening hemoptysis. The remaining 510 patients with pulmonary hydatid cyst were considered as group I. At presentation, chest radiography was available in 95% of patients. Data were collected from the hospital charts were analyzed in terms of age, radiological findings, symptoms and signs, and management. Patients with massive hemoptysis due to other reasons or mild hemoptysis were excluded from the study. Emergency posterolateral thoracotomy was the most commonly used surgical procedure (10 patients); in two of these ten patients, anterior thoracotomy was also performed. After extraction of the inner cyst membrane, lobectomy of the destroyed lung was performed in nine patients. In the remaining patients, suturing of the bronchial opening was carried out primarily followed by lobectomy due to the continuation of air leak. Unfortunately, two patients died due to blood aspiration and adult respiratory distress syndrome. In the postoperative period, albendazol (10 mg/Kg per day) was given to all patients for at least 3 months to prevent recurrence of the disease. The mean follow-up period was 2.4 ± 0.85 years (range 6 months to 5 years). In group I, 510 patients with pulmonary hydatid cyst were operated on and compared with group II. There were (n=284) 56% men and (n=226) 44% women with a mean age of 33.51 ± 8.66 years (range 3 to 76 years).

Statistical analyses

Data analysis included Chi-square and Fisher's exact tests for categorical variables. A p-value of <0.05 was considered significant. All analyses were performed using SPSS software (version 14.0).

RESULTS

Demographic features of groups I and II are shown in Table 1. There were no significant differences in age ($\chi 2= 1.19$, df=1, p =0.27) and sex between the two groups ($\chi 2= 0.97$, df=1, p = 0.32). Symptoms upon presentation of patients with life-threatening hemoptysis (group II) and important symptoms of group I on admission are summarized in Table 2. All ten patients of group II had presenting symptoms of cough, massive hemoptysis, and respiratory distress. Four of ten patients with massive

hemoptysis had a history of minor or mild hemoptysis and were diagnosed based on a radiographic study. Seven of ten patients were asymptomatic, and hydatid cyst was diagnosed incidentally.

Table 1. Demographic features of 520 patients with pulmonary hydatid cyst

Age/ Sex		Patients. N(%) Group 1 N=510	Group 2 N=10	X^2	df	p-value
Age	< 12 years old	49 (9.6)	2 (20)	1.19	1	0.274
	> 12 years old	461 (90.4)	8 (80)			
Sex	Female	226 (44)	6 (60)	0.97	1	0.320
	Male	284 (56)	4 (40)			

Table 2. Important symptoms of pulmonary hydatid cyst in 520 patients on admission

Symptom	Patients. N (%) Group1 n=510	Group2 n=10	X^2	df	P-Value
Cough	471 (92.4)	10 (100)	0.82	1	0.363
Massive hemoptysis	0 (0)	10 (100)	520	1	<0.0005
Respiratory distress	24 (4.7)	10 (100)	145.7	1	<0.0005
Chest pain	369 (72.4)	9 (90)	0.154	1	0.215

All patients had chest X-rays as part of their preoperative workup. Radiological findings of all patients are shown in Table 3.

Table 3. Radiological findings in 520 patients with pulmonary hydatid cyst.

Radiologic Findings	Patients. N (%) Group1 n=510	Group2 n=10	X^2	df	P-Value
Water Lilly sign	79 (15.5)	2 (20)	0.152	1	0.697
Air fluid level	92 (18)	1 (10)	0.432	1	0.511
One lung infiltrate	0(0)	1 (10)	51.09	1	0.019
Bilateral lung infiltrate	0(0)	2 (20)	102.39	1	<0.0005
Cavitary image	0(0)	2 (20)	102.39	1	<0.0005
Ruptured giant hydatid cyst	0(0)	2 (20)	102.39	1	<0.0005
Unruptured hydatid cyst	258 (50.6)	2 (20)	3.67	1	0.055
Ruptured hydatid cyst	252 (49.4)	8 (80)	3.68	1	0.055
Notch Sign	242(47.5)	2 (20)	2.96	1	0.085

A definite diagnosis was established in seven patients (70%) following a chest x-ray examination. The diagnosis of the remaining three patients was achieved following thoracic operations. Radiological findings were also compared between the two groups. The type of operation for all patients is listed in Table 4.

Table 4. Principle operation on 520 patients with pulmonary hydatid cyst.

Patients	Patients. N (%) Group1 n=510	Group2 n=10	X^2	df	p-value
Open lobectomy	10(2)	10(100)	254.9	1	<0.0005
Lobectomy & Decortication	22(4.3)	2(20)	5.48	1	0.019
Evacuation of LM* and suture of bronchovascular opening	452(88.6)	1(10)	54.01	1	<0.0005
Emergency thoracotomy	0(0)	10(100)	520	1	<0.0005
Segmentectomy	26(5.1)	0(0)	0.537	1	0.464

*LM= Laminated membrane

According to the results obtained from post-surgical pathological examination, all patients had pulmonary hydatid cysts. Six patients (60%) had hydatid cyst in the right lung and four (40%) in the left lung. Three patients had infected hydatid cysts leading to suppuration. Summary of the patients' details are shown in Table 5.

In group II, postoperative complications included immediate air leak (n=2, 20%), atelectasis (n=6, 60%), and respiratory distress syndrome (n=2, 20%). Eight patients had accompanying hydatid cyst of the liver. An additional postoperative workup (chest x-ray, ultrasonography, and CT-scan of the chest) showed no recurrence in the follow up period. Two patients needed ventilatory support and died due to blood aspiration and acute respiratory distress syndrome, one on the third and the other on the fourth day after operation. All patients received albendazol 10 mg/kg per day immediately after operation.

Table 5. Summary of pulmonary hydatid cyst patients' details (group II).

Patient No.	Sex, Age	Volume of Massive Hemoptysis (cc)	Radiology	Involved Lobe	Surgery	Outcome
1	F/8	1200-1300	Cavitary Ruptured giant HC	LLL	a,b,c	Death (BA,ARDS)
2	M/46	1300-1400	Infiltration Water lily ruptured	RLL	b,c	Successful
3	F/46	900-1000	Infiltration ruptured	RML	b,c	Death (BA,ARDS)
4	F/23	900-1000	Water lily Ruptured	RLL	b,c	Successful
5	F/35	600-700	Notch sign Intact	RML	b,c	Successful
6	F/40	600-700	Air fluid level Ruptured	RLL	b,c	Successful
7	M/28	700-800	Water lily Ruptured	LLL	b,c	Successful
8	M/18	600-700	Notch sign Intact	RLL	b,c	Successful
9	F/26	800-900	Infiltration Ruptured	Lingula	b,c	Successful
10	M/9	700-800	Giant hydatid cyst Ruptured	LLL	b,c	Successful

Surgery (a): Thoracotomy & evacuation of laminated membrane & suture closure of bronchovascular openings.

Surgery (b): Thoracotomy & evacuation of laminated membrane & lobectomy

Surgery (c): Emergency thoracotomy

HC= Hydatid Cyst BA= Blood Aspiration ARDS= Adult respiratory distress syndrom

The patients were also evaluated in terms of potential risk factors for the disease. Six patients were living in rural areas and had contamination with dogs and sheep. No correlation was found between life-threatening hemoptysis and intact hydatid cyst (χ^2=2.25, df=1, p= 0.094), rupture of the cyst (χ^2=3.67, df=1, p= 0.055), and infection (χ^2=0.609, df=1, p= 0.435). However, no significant difference was found between life threatening hemoptysis and size of the cyst (χ^2=0.068, df=1, p= 0.794).

Data analysis of complications in group I and group II with chi- square test showed 4.1 % versus 20% respectively. This result showed a significant difference between the two groups (χ^2= 5.82, df=1, p< 0.16).

Two patients in group II died whereas no mortality occurred in group I. Fisher's exact test showed a significant difference in the mortality rate between the two groups (χ^2= 102.39, df=1, p<0.0005).

Figure 1 shows Kaplan – Mayer curve of overall survival in group II.

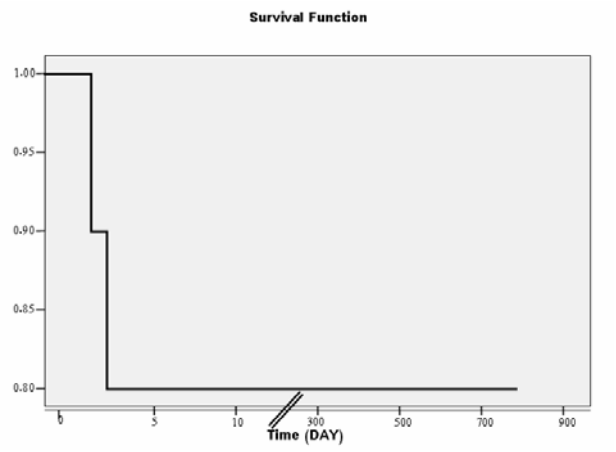

Figure 1. Kaplan – Mayer curve of overall survival of group II

DISCUSSION

Hydatidosis is widespread in developing countries because of inadequate hygiene and mainly affects young adults (6). Hydatid cyst is a rare cause of massive hemoptysis. Cough, acute respiratory

distress, hemoptysis and chest pain are the most common presenting symptoms. The incidence of hemoptysis is 38% in children and up to 70% in adults (7). The incidence of massive hemoptysis obtained in the present study was 1.9% which was higher than 0.0-0.9% value reported by others (3,5). Hemoptysis in adults is most often caused by tuberculosis, bronchitis, bronchiectasis, trauma, or bronchogenic carcinoma. Pediatric hemoptysis is most likely secondary to infection, tracheostomy-related problems or foreign body aspiration (7). Cystic echinococcosis has also been a cause of hemoptysis. Bronchial fistulation and rupture is an important event in the process of cyst development. This complication leads to hydatid vomica, hemoptysis, and cyst infection. At the time of diagnosis, rupture in bronchial tree is observed in one third of the patients. Hydatidoptosis is a specific sign and consists of evacuation of entire small hydatids by vomica. Another cause of massive hemoptysis relates to erosion of pulmonary hydatid cyst to the thoracic aorta (8). The mechanism of hemoptysis in pulmonary hydatid disease may be due to erosion of a bronchus or obstructive effect of bronchial infection. This may lead to further erosion of bronchial arterial supply leading to hemoptysis. Massive hemoptysis is a potentially lethal condition that needs to be investigated thoroughly and brought under control promptly. The mortality rate depends mainly on the underlying etiology and the magnitude of bleeding (8, 9). Although the diagnosis of hemoptysis may be established by chest radiography, the pathology may be missed. Complicated cases may have atypical radiological presentations when it appears heterogeneous or is associated with bronchial obliteration and atelectasis (8). Rupture may change the radiological appearance of the cyst, resulting in a wrong diagnosis. Finding the etiology of hemoptysis is important. Urgent bronchoscopy should be performed in unstable patients because it has an important role in diagnosis and therapy. (4,10). Bronchoscopy and CT-scan may show pathologies not detectable by chest radiography. Serological tests are less sensitive in patients with lung hydatidosis than in those with liver location (8,11). It may be used to facilitate the introduction of balloon tip catheters into the bleeding bronchus for tamponade of the hemorrhagic artery, protecting the controlateral lung or non-bleeding bronchi from blood aspiration. Bronchial tamponade should only be used as a temporary measure until definitive treatment is instituted. Bronchial artery embolization is an excellent nonsurgical alternative method suited for patients unfit for anesthesia (4). There is no obvious reason why intact cysts should bleed at all, but the mechanism of intact hydatid cyst may be explained on the basis of bronchiolar arterial hypertension (12).

Surgery is recommended urgently as the treatment of choice in massive hemoptysis due to complicated pulmonary hydatid disease. Most authors believe that complicated cysts require more extensive procedures than uncomplicated cysts (13). Our principle indications for lobectomy were severe hemorrhage with ruptured giant hydatid cyst (20%), large pulmonary hydatid cyst that destroyed more than 50% of the lung in 30% of patients, suppuration of the cystic cavity (20%), no salvageable lobar parenchyma (40%), and concomitant aspiration with stiffness of the lobe in 30% of patients.

The diagnosis in our patients was only clinical-radiological and was not supported by serology or therapeutic response to albendazol or bronchoscopy. In cases of pulmonary hydatidosis, the clinical and radiological picture was so unique that it could be easily recognized despite its rarity.

The mortality and morbidity of surgical treatment

of massive hemoptysis due to pulmonary hydatid cysts are high. In our study, mortality was 20% which was due to aspiration of blood and acute respiratory syndrome. Immediate air leak and atelectasis were the most common complications. The postoperative hospital stay ranged between 7 and 25 (mean 11.30 ± 6.56) days. In our series, 2.5 years follow-up revealed no recurrences.

CONCLUSION

Hydatidosis is the most frequent pulmonary parasitic disease. Massive hemoptysis is one of the most critical complications that rarely occurs. Our study stresses emergency surgery as an elective and effective treatment for massive hemoptysis from pulmonary hydatid cysts. A ruptured hydatid cyst with massive hemoptysis is a life-threatening condition associated with increased mortality and morbidity, more extensive surgery and longer hospitalization periods. Such patients require ICU care for longer periods of time and ventilatory support.

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