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Original Article

Fifteen Years Experience with Pulmonary Hydatidosis in Zahedan, Iran

***B Sharifi-Mood [1], A Fazaeli [1], Sh Izadi [1], S Mokhtari [2]**

[1]Research Center for Infectious Diseases and Tropical Medicine, Zahedan University of Medical Sciences, Zahedan, Iran
[2]Boo-Ali Center for Diseases Control and Prevention, Zahedan, Iran

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Abstract

Background: Hydatid disease is a major world health problem and pulmonary hydatidosis is a widespread disease. It is presented with different clinical manifestations. In order to determine the most clinical manifestation, diagnostic methods and clinical outcome in our patients, we conducted this study.

Methods: Forty-nine patients with pulmonary hydatid cysts who were admitted to our hospital in Zahedan (Southeast of Iran) between 1990 and 2005, evaluated. We retrospectively reviewed the patients' symptomatology, diagnostic studies, treatment options, and morbidity as well as mortality rate.

Results: The ages of the patients ranged from 16 to 68 years (mean 43 years). Seventy–five percent of patients were from male gender. Hemoptysis was one of the most common clinical presentations in our patients. Radiological studies were the main diagnostic tool. The correct preoperative diagnosis was made in 92% of the patients by chest roentgenogram plus chest CT-Scan. Eighty seven percent of patients were treated by surgical route. Only one patient was expired during surgery.

Conclusion: Upon the results emerged from this study, hemoptysis is one of the most prevalent clinical manifestation in patients with pulmonary hydatidosis and it can mimic pulmonary tuberculosis in endemic area.

Keywords: *Pulmonary hydatid cyst, Hemoptysis, Surgery, Albendazole, Iran*

Introduction

Echinococcosis or hydatid disease is caused by larvae, which is the metacestode stage of the tapeworm *Echinococcus* (1). Of the 4 known species of *Echinococcus,* 3 are of medical importance in humans (2). These are *E. granulosus,* causing cystic echinococcosis (CE); *E. multilocularis,* causing alveolar echinococcosis (AE); and *E. vogeli*. *E. granulosus* is the most common of the rest. *E multilocularis* is rare but is the most virulent, and *E. vogeli* is the most rare (2, 3).

The incidence of CE in endemic areas ranges from 1 to 220 cases per 100,000 inhabitants, while the incidence of AE ranges from 0.03 to 1.2 cases per 100,000 inhabitants, making it a much more rare form of echinococcosis (2, 3). Hydatid disease is found worldwide and considerable public health problems are encountered in endemic areas (3-5). Hydatid disease is endemic in rural areas in many countries, especially in countries around the Mediterranean and those in the Middle East, Central Asia, South America, South Africa, New Zealand and Australia (6-10). Hydatid cysts most often affect the liver (68.8%–75.2% of cases) but also affect the lungs (17.2%–22.4%) (7). Cough, chest pain, and breathlessness are the most common presenting symptoms in pulmonary hydatidosis and hemoptysis sometimes occurs (2, 10-12). Rupture of the cyst and infection are the most common complications.

***Corresponding author**: Tel: +98/541 3236969 Fax: +98-541 3212975 E-mail: batoolsharifi@yahoo.com

Data on the epidemiology of pulmonary hydatid cyst in Southeast of Iran is unclear and the disease is still an important public health problem. In this article, we present and discuss our experience of fifteen years with respect to clinical presentation, diagnostic tools and the treatment of pulmonary hydatid disease.

Materials and Methods

In this retrospective study, we reviewed a series of patients with pulmonary hydatid disease who were admitted to infectious wards in Boo-Ali Hospital, during a period of fifteen years, from April 1990 to February 2005. Boo-Ali Hospital is a teaching hospital in Zahedan in Sistan and Baluchistan Province and a referral center for infectious diseases and tropical medicine located in Southeast of Iran. First, we selected all of the patients with hydatid disease and then evaluated the patients with pulmonary hydatidosis. All patient files were reviewed retrospectively for presenting symptoms, diagnostic studies, kind of treatment, complications and mortality**.**

Results

Forty–nine patients (37 males, 12 females; mean age 43; age range: 16 to 68 years) with pulmonary hydatidosis were evaluated. Thirty nine of the patients (81%) were from the rural areas of the Sistan and Baluchistan Province. All patients (100%) were symptomatic. Coughing (73%) and hemoptysis (65%) were the most frequent symptoms at presentation time. Recurrent hemoptysis was found in 58% of patients but it was not severe. Fever, chest pain, purulent sputum, weakness and dyspnea were other frequent symptoms of the cystic disease. No anaphylactic reaction was observed (Table1). The diagnosis was based on chest X-ray in 69% of patients and in 92% of patients by chest roentgenogram plus chest CT-Scan. Diagnosis was made in four patients during surgery. These patients were primarily treated medically as pulmonary abscess and when they did not respond to medical therapy referred to surgeon. Pathologic findings confirmed the pulmonary hydatid cyst. Serological studies (ELISA) were done in only18 patients and confirmed the diagnosis. The right lower lobe was the site most frequently involved (27%) (Table 2). The disease was unilateral in 45 of the patients (92%) and multiple cysts were present in 4 (8%). Two cases had pulmonary and extrapulmonary cysts (liver) .Ten (20.4%) of the cysts was infected and one patient was referred to our hospital with empyema. Eighty- seven percent of patients were treated by surgical interventions and resection of the cyst was the most common procedure (76%). Radical surgical procedures such as lobectomy (3 cases) or segmentectomy (2 cases) was performed in 11% of the patients due to large cyst & damaged lung parenchyma. Six patients (12.4%) were treated only by medical route & albendazole was used in elderly patients who could not tolerate surgery. All operated patients were received albendazole 3 days before & one month after the operation. The patients did not closely follow-up to evaluate the success of therapy & adverse effects such as neutropenia, liver dysfunction especially when they were treated medically because the most of the patients were from the rural areas. Five patients had a previous history of pulmonary hydatidosis. All cysts were in the same operated site & all patients were treated with a second thoracotomy & albendazole. Only one patient expired due to massive hemorrhage during surgery.

Table 1: Frequency of clinical manifestations in the patients with pulmonary hydatidosis

Clinical sign	No.	Percent
Cough	36	73.8
Hemoptysis	32	65.3
Fever	27	55
Chest pain	18	36.7
Purulent Sputum	17	34.6
Dyspnea	16	32.6
Weakness	10	20.4

Table 2: Frequency of Pulmonary involvement according to lobe involvement

Site involvement	No.	%
Right upper lobe	7	13.7
Right middle lobe	12	23.5
Right lower lobe	14	27.4
Left upper lobe	10	19.6
Left lower lobe	8	15.6

Discussion

Hydatid disease is a major world health problem and pulmonary hydatid cyst is a common form of hydatidosis. Cough, fever and chest pain are common presenting symptoms in pulmonary hydatidosis but hemoptysis can occurs (2, 12). While hemoptysis is a presenting symptom in adult patients in up to 70%, it is a rare presentation in pediatric patients (12). The mechanism of hemoptysis in pulmonary hydatid disease may be due to pressure erosion of a bronchus or obstructive effect with bronchial infection. This may in turn lead to further erosion into a branch arterial supply leading to hemoptysis. Hydatid cysts may also erode vascular structures including the aorta causing massive hemoptysis (13). Therefore, in endemic area where tuberculosis is also prevalent, it can mimic pulmonary TB especially, when they refer with hemoptysis. In pulmonary hydatidosis, cysts are predominantly located in the lower lobes & more on the right side than on the left (13). In our patients, 27% of the cysts were in the right lower lobe and the disease was unilateral in 92% of the patients. Our results were also in accord with previous reports (1, 4, 6, 9). In a Turkish study, the right lower lobe was the site most frequently involved (26%) and the disease was unilateral in 67 of the patients (96%) (6). In the present study, hemoptysis was more common than previous studies (1, 4, 6, 9). All cases with hemoptysis had an age more than 20 years. Singh *et al*. presented a ten-year old child with recurrent hemoptysis and a bout of massive hemoptysis due to pulmonary hydatidosis (12). The combination of imaging and serology usually enables diagnosis. The plain chest radiography is the main diagnostic tool in hydatid disease (4). It has been shown to be highly sensitive (90-98%) in several studies (13, 14). Typically, intact pulmonary cysts are seen as well demarcated, spherical, homogenous and single or multiple lesions (14). Some characteristic descriptive images of hydatid cysts such as the water-lily

sign, Escudero-Nenerow sign, notch sign, double dome arch sign, meniscus sign, air-bubble sign have been reported. However, plain chest x-ray is not always enough for the diagnosis (12). Computed tomography of the chest, by revealing the fluid density of a cystic lesion, the air fluid density of a cavitary lesion, or the solid density of a complicated cyst, may be helpful in establishing the diagnosis (14, 15). It can also be of value in determining the presence of cysts in areas difficult to visualize with chest x-ray films, especially in the posterior and costophrenic angles (13-17). Ninety two percent of our cases could be detected preoperatively by chest x-ray plus CT-Scan. In Yeginsu study, also the correct preoperative diagnosis could be made in 71% of the patients by plain chest roentgenogram and in 96% of the patients by chest roentgenogram plus tomography (6).

Serological studies are highly sensitive (70-100%) in hydatid disease, but in some reports false-negative results are reported in 30% of the cases (6). Additionally, false positivity in some parasitic, degenerative, autoimmune, allergic, and neoplastic diseases is reported (6, 13, 14). Serological studies had been confirmed the diagnosis in eighteen of our patients.

The treatment of pulmonary hydatid disease is surgical. Lung-preserving surgical interventions are the treatment of choice (18). Choice of surgical method depends on the condition of the cysts. Cystotomy and capitonnage is the most common procedure and is preferred by many teams (5, 6, 14, 19-23). Medical therapy generally should begin ≥4 days prior to surgery and be continued for 1–3 months (4, 12, 15, 23). Albendazole (10–15 mg/kg body weight orally in two divided doses, usually 800 mg daily) is used (16-18). Albendazole is preferred because it has better bioavailability than mebendazole (6,19).Two study by Keshmiri *et al*. showed that albendazole has good effect on hydatid cysts and should be offered to patients before surgical treatment is considered (20, 21). Eighty-seven of our patients received surgical preservative procedures. Radical surgery was performed in 11% of cases. However, high rates of radical surgery were reported in other series (6, 18). Video-assisted thoracoscopic and percutaneous aspiration techniques may be performed in selected patients but have not been widely accepted yet (6). Recent method was performed in only one of our patients. Surgical treatment of pulmonary hydatid disease carries a morbidity risk of 0 to 17% and a mortality risk of 0 to 5%, while recurrence of cysts varies from 2 to 30% over 5 years even without obvious spillage of cyst contents (4, 10). In our study, only one patient was expired due to massive hemorrhage & we could not follow-up our patients to evaluate the success of therapy and for adverse effects.

In conclusion, hemoptysis is one of the most prevalent clinical manifestations in patients with pulmonary hydatidosis in Southeast of Iran. Since, tuberculosis is also endemic in this area, it can mimics pulmonary tuberculosis. Therefore, clinician should be aware to pulmonary hydatidosis when he or she visit a patient with hemoptysis.

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The authors declare that they have no Conflict of Interests.

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