Primary Pulmonary Echinococcosis-A Case Report

Md Rafiqul Alam[1], Md Abul Kalam Azad[1] , Zia Uddin Ahmed[2] , Md Mizanur Rahman Khan[3], Ferdous Jahan[3], Hasan Imam[3], Shah Md. Mohaimenul Haq[4], Nur Mohammad[4].

[1]Associate Professor, [2]Resident, [3]Medical Officer, [4]Resident, Dept of Internal Medicine, BSMMU.

Abstract:

Echinococcosis is a parasitic disease endemic in many parts of the world. Liver is the most common affected organ followed by lungs. Primary pulmonary involvement is very rare. In this case report, we are going to present a case of primary pulmonary echinococcosis in a middle age female. She was presented with cough and haemoptysis. She was initially labeled and treated as a case of consolidation with different antibiotics in appropriate dose and duration without clinical and radiological improvement. So she was evaluated further and diagnosed as a case of primary pulmonary echinococcosis on the basis of histopathology and positive serology for Echinococcal antigen. She was advised to take oral Albendazole 400mg 12 hourly for one year with regular follow-up.

Keywords: Pulmonary Echinococcosis, Hydatidosis, Albendazole

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Introduction:

Echinococcosis, also known as hydatidosis is one of the most important zoonotic diseases in the world. The endemic areas are the Mediterranean countries, the Middle East, the southern part of South America, Iceland, Australia, New Zealand, and southern parts of Africa; the latter 5 are intensive endemic areas. Central Asia, particularly China, is also an endemic area[1,2]. The incidence of cystic echinococcosis (CE) in endemic areas ranges from 1-220 cases per 100,000 inhabitants, while the incidence of alveolar echinococcosis (AE) ranges from 0.03-1.2 cases per 100,000 inhabitants, making it a much more rare form of echinococcosis[3].We report one of the such cases of primary pulmonary echinococcosis in a middle age female at our setting.

Case Summary:

A 35-years-widow female, DOTS medicine provider hailing from Bogra presented with the complaint of cough for 1 year, haemoptysis for last 2 months. Cough was dry, persistent, no diurnal & seasonal variation with occasional haemoptysis for last 2 months. She had no history of fever, weight loss, chest pain or breathlessness . She was initially evaluated on the basis of chest skiagram (fig- 1) and was labeled as a case of consolidation. She was treated with different antibiotics in adequate dose and duration, without any clinical and radiological improvement. For proper evaluation and management, she was admitted in Bangabandhu Sheikh Mujib Medical University on 20/06/2015. She was a mother of 5 children with good health. Her husband and father died due to cerebrovascular accident. She was of poor personal hygiene and used to walk bare footed in home. She had pet sheep and dog. On examination, she was afebrile and anaemic. Her pulse was 90/min, BP-120/70 mmHg, Respiratory rate-18/min. Respiratory system examination revealed dullness on percussion with diminished breath sound on right 3rd to 5th intercostals space along right mid clavicular line.

On admission, her Hb was 11.6 gm/dl, TC-9,000/mm3 with normal differential and ESR was 39 mm in 1st hour. Chest X-ray P/A view shows two thick wall cavitary lesions, one in the right and another on left lower zone with surrounding non homogenous opacities. (Fig-2).

Corresponding Author: Dr. Rafiqul Alam, Associate Professor
Department of Internal Medicine, BSMMU.
Email:rafiq.bsmmu@gmail.com

Sputum for AFB ,Gene Xpert, Gram stain- were negative.

CT finding shows diffuse air-space densifications with multifocal streaky fibrosis and one thin walled cavitary lesion which are noted in right upper lobe. A thin walled cavitary lesion having multiple air pockets within and surrounding hyper attenuated area is noted in the apical and lateral basal segment of right lower lobe. Another one smaller cavitary lesion is also noted in apical and medial basal segment of left lower lobe which is more suggestive Koch's lesion with multiple tuberculomas and pleural thickening in above mentioned areas. (Fig-3 & 4)

CT guided FNAC was done from lung parenchymal lesion.The histopathology shows adequate cellular material containing a good number of polymorph, histocytes and few number of lymphocyte with mixed degenerated scoles material and hyalinized. No malignant cell or granuloma is seen which are compatible with Hydatid disease (Infective) (Fig-5,6).
Echinococcal antigen was found positive.
USG of whole abdomen was normal.

A diagnosis of primary pulmonary echinococcosis was made on the basis on physical examination and investigation. She was treated with albendazole 400 mg twice daily (10-15mg/kg/day) for one year [4,5][Table 1] .We advised the patient follow up with CBC count and liver enzyme evaluation at biweekly intervals for 3 months and then every 4 weeks to monitor for toxicity.We also adviced ELISA or indirect hemagglutination tests are at 3, 6, 12, and 24 month intervals and ultrasonography and/or CT scan at the same intervals as the laboratory tests[6]

Table-1

Drugs used in the treatment of echinococcosis

	Oral Dosage	Duration	Maximum dose
Mebendazole	40-50mg.kg body weight day-1 three times daily	3-6 month for E. granulosus	6g.day-1
Albendazole#	10–15 mg•kg body weight-1 •day-1twice	3-6 month for E. granulosus and prolonged	Usually 800 mg. day-1
	daily for E.multilocularis	or lifelong	
Praziquantel*	40 mg.kg body weight-1 once weekly	Uncertain	NA

• NA: not available
• #: preferred because it exhibits better bioavailability than mebendazole
• *: can combine with albendazole

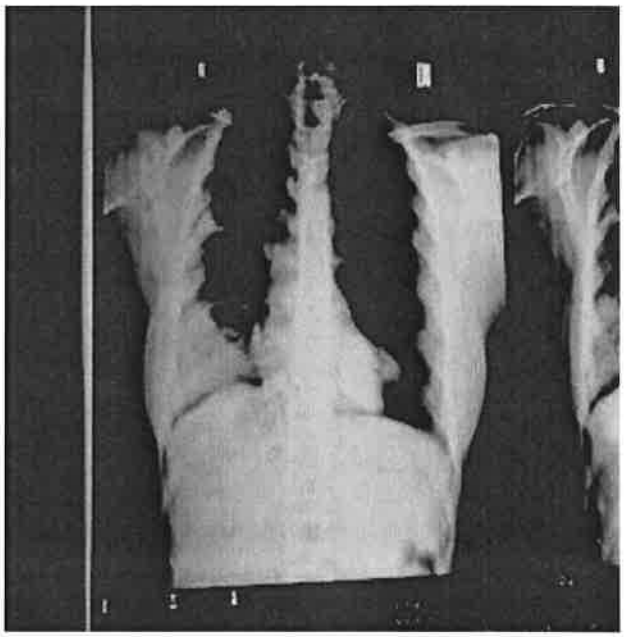

Fig-1: (On 20/05/15)

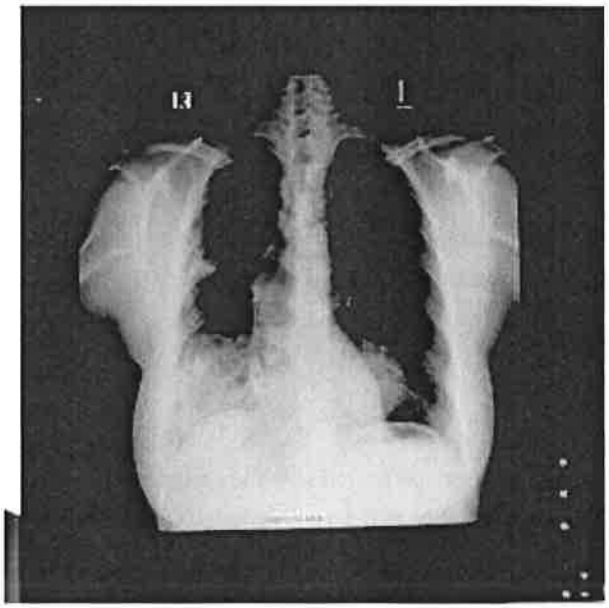

Fig-2: (On 08/06/15)

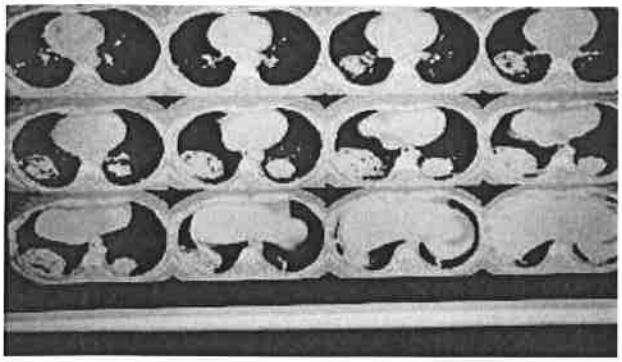

Fig-3:

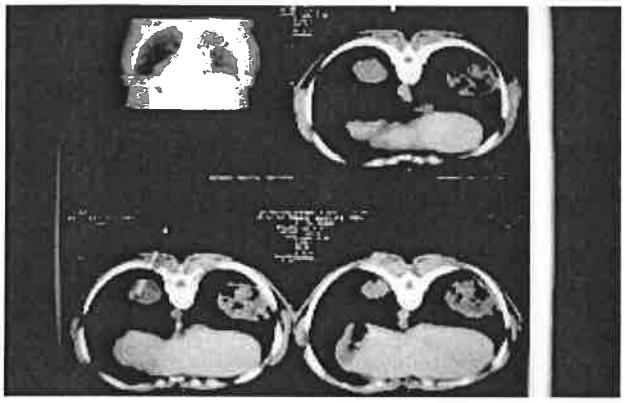

Fig-4:

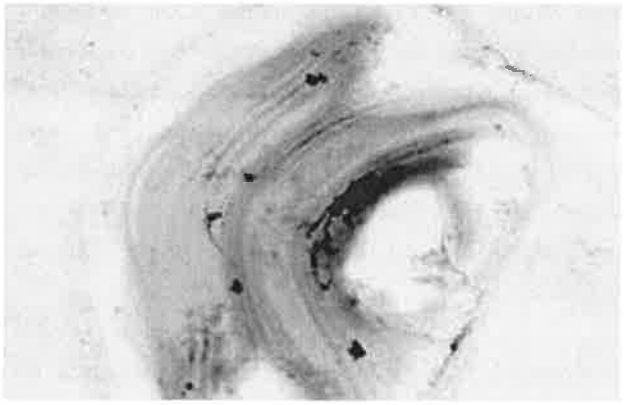

Fig-5:

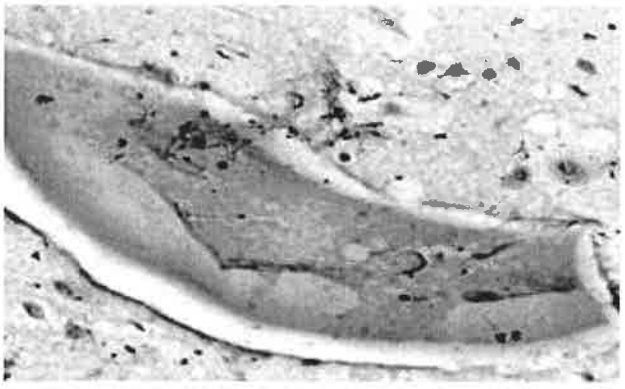

Fig-6:

Discussion:

Echinococcosis or hydatid disease is caused by larvae, which are the metacestode stage of the tapeworm Echinococcus. Four species are recognized and belong to the family Taeniidae. The vast majority of infestations in humans are caused by E. granulosus. E. granulosus causes cystic echinococcosis, the pastoral form, which has a worldwide distribution and is concentrated in sheep-raising areas. Humans are exposed less frequently to E. multilocularis, which causes alveolar echinococcosis, because E. multilocularis infestation usually occurs in colder areas and is associated with animals in wild ecosystems, especially foxes. E. vogeli and E. oligarthrus are rare species and cause polycystic echinococcosis.[7]

Humans contract the disease accidently from water or food or by direct contact with dogs. The organism can reach the lung in many ways-either secondarily during circulation after it crosses the liver, via lymphatic vessels bypassing the liver, following intrathoracic rupture of a cyst of the liver, or by inhalation of the eggs causing primary lung disease.[7] The liver is the most common site of cyst formation, followed by the lung in 10–30% of cases and other sites (usually the spleen, kidney, orbit, heart, brain and bone) in ⊠10% of cases.[7] In children, the lungs may be the commonest site of cyst formation.[8] Of patients with lung cysts, ⊠20–40% also have liver cysts [9,10]. Pulmonary hydatid disease affects the right lung in ⊠60% of cases, 30% exhibit multiple pulmonary cysts, 20% bilateral cysts and 60% are located in the lower lobes.[8] Within the chest, echinococcosis can primarily involve the pleural cavity mediastinum and chest wall.[8] The initial phase of primary infection is asymptomatic and may remain so for many years. Hydatid disease is seen in subjects of any age and sex, although it is more common in those aged 20–40 yrs.[10] Most intact lung cysts are discovered incidentally on chest radiographs. Subsequent clinical features of E. granulosus infection depend upon the cyst site and size. Small cysts may remain asymptomatic indefinitely, but large cysts may cause symptoms by compressing adjacent structures. Mediastinal cysts may erode into adjacent structures causing bone pain, haemorrhage or airflow limitation. Symptomatic hydatid disease of the lung, however, more often follows rupture of the

cyst. Rupture may be associated with the sudden onset of cough and fever.

The most valuable diagnostic method in pulmonary hydatid disease is the plain chest radiograph.[10] Pulmonary cysts may range between 1 and 20 cm in diameter.[11] Computerised tomography (CT) scan with contrast may demonstrate a thin enhancing rim if the cyst is intact . The contents of closed simple cysts are homogeneous, with a density close to that of water.[12] The magnetic resonance signal characteristics of a hydatid cyst may differ depending on the developmental phase, i.e. whether it is uni- or multilocular and whether the cyst is viable, infected or dead. Information regarding reactive changes in the host tissue, capsule and signal intensity of parent and daughter cysts is also obtained. On magnetic resonance imaging (MRI), cysts show low signal intensity on T1-weighted images and high signal intensity on T2-weighted images.[13]

In pulmonary cystic echinococcosis, routine laboratory tests do not show specific results. Less than 15% of cases exhibit eosinophilia, which generally occurs only if there is leakage of antigenic material.[10] An enzyme-linked immunosorbent assay or indirect haemagglutination test is commonly used as an initial screen, and is positive in only ⊠50% of patients with pulmonary hydatidosis and >90% of patients with hepatic cysts.[14] Currently, polymerase chain reaction (PCR) techniques are only being used for research purposes, but they may have a role to play in diagnosis and species determination in the future. Deoxyribonucleic acid probes using Southern hybridisation tests are also being developed.

Patients who are able to undergo surgery, it is considered the treatment of choice since the parasite can be completely removed and the patient cured. The surgical options for lung cysts include lobectomy, wedge resection, pericystectomy, intact endocystectomy and capitonnage.[15] Puncture, aspiration or injection of a helminthicide and reaspiration has been advocated for hepatic cysts. However, for pulmonary cysts, this technique shows more complications and is rarely indicated.[16,17] Bilateral hydatid disease of the lungs may be managed by one- or two-stage surgery via either bilateral thoracotomy, sternotomy or video-assisted thoracic surgery. Some prefer two-stage thoracotomy, operating on the side with the larger ruptured and infected cyst first. However, median sternotomy is a better alternative for the treatment of bilateral hydatid disease of the lung. This method is more economical, causes less pain and is better tolerated than two thoracotomic procedures.[4,14,18-20]

Medical therapy with benzimidazoles is valuable in disseminated disease, including secondary lung or pleural hydatidosis, poor surgical risk patients and when there is intraoperative spillage of hydatid fluid (table 1).[17] The usual dose of mebendazole is 40–50 mg/kg body weight per day, given in three divided doses after meals (maximum daily dose 6g). Therapy is usually indicated for o3–6 months. Albendazole is given at a dosage of 10–15 mg/kg body weight per day in two divided doses and the usual dose is 800 mg daily. Therapy is most often indicated for a minimum of 3–6 months. Albendazole is preferred because it has better bioavailability.[5]

Both drugs are also contraindicated in pregnancy (especially during the first trimester) because of possible teratogenicity. Medical treatment alone has been suggested by some to be sufficient for small pulmonary hydatid cysts . A newer benzimidazole compound, oxfendazole, has been studied in a mouse model and preliminary results suggest it may be a more effective compound.[21]

Praziquantel, an isoquinolone, has also been used for therapy. It has been shown to have effective protoscolicidal activity and may be more effective than albendazole in vitro.Praziquantel (40 mg/kg body weight orally once a week) has been used alone and in combination with albendazole. A few reports suggest that the combination of albendazole and praziquantel as medical therapy or as postspillage prophylaxis is more effective than either therapy alone.[14]

Evaluating the success of therapy is difficult and usually requires regular follow-up and imaging. The use of serological titres to monitor therapy has also been assessed.[6] Serological tests showed an increase in titre in

the majority of patients for the first 3 months after surgery, probably a result of antigen liberation during cyst manipulation. Serological tests showed decreasing antibody titres from 3 months after surgery in patients without relapse. Patients who relapse show either persistently high (early relapse) or an initial decrease and subsequent increase (late relapse) in antibody titres.[6]

Prevention of cystic echinococcosis can often be achieved by avoiding close contact with dogs. Careful washing of fresh produce can also reduce infection. Prohibition of home slaughter of sheep and proper offal disposal prevents dogs from consuming infected viscera, thus disrupting the life cycle of the parasite. Elimination of stray dogs and surveillance techniques, involving either diagnostic purging of dogs or coproantigen tests, have helped to reduce infections in some endemic areas . Vaccination is also a prospect for prevention of echinococcosis, since protective immunity develops in intermediate hosts.

References:

01. Wang K, Zhang X, Jin Z, Ma H, Teng Z, Wang L. Modelling and analysis of the transmission of echinococcosis with application to Xinjiang Uygur Autonomous Region of China. J Theor Biol. 2013;10.

02. Torgerson PR.The emergence of echinococcosis in central Asia. Parasitology. 2013;10:1-7.

03. Moldovan R, Neghina AM, Calma CL, Marincu I, Neghina R. Human cystic echinococcosis in two south-western and central-western Romanian counties: A 7-year epidemiological and clinical overview. Acta Trop. 2012;121(1):26-9.

04. Gottstein B, Reichen J. Hydatid lung disease (echinococcosis/hydatidosis). Clin Chest Med 2002;23:397–408. Gottstein B, Reichen J. Hydatid lung disease (echinococcosis/hydatidosis). Clin Chest Med 2002; 23:397–408.

05. Anadol D, Ozcelik U, Kiper N, Gocmen A. Treatment of hydatid disease. Paediatr Drugs 2001;3:123–35.

06. Zarzosa MP, Orduna Domingo A, Gutierrez P. Evaluation of six serological tests in diagnosis and postoperative control of pulmonary hydatid disease patients. Diagn Microbiol Infect Dis 1999; 35:255–62.

07. Schantz P. Echinococcosis. In: Guerrant R, Walker DH, Weller PF, eds. Tropical Infectious Diseases: Principles, Pathogens and Practice. Philadelphia, WB Saunders, 1999;1005–25.

08. Thumler J, Munoz A. Pulmonary and hepatic echinococcosis in children. Pediatr Radiol 1978; 7: 164–71.

09. Gomez R, Moreno E, Loinaz C. Diaphragmatic of transdiaphragmatic thoracic involvement in hepatic hydatid disease: surgical trends and classification. World J Surg 1995;19: 714–19.

10. Dogan R, Yuksel M, Cetin G, et. Surgical treatment of hydatid cysts of the lung: report on 1055 patients. Thorax 1989; 44: 192–99.

11. Saksouk FA, Fahl MH, Rizk GK. Computed tomography of pulmonary hydatid disease. J Comput Assist Tomogr 1986; 10: 226–32.

12. Singh S, Gibikote SV. Magnetic resonance imaging signal characteristics in hydatid cysts. Australas Radiol 2001; 45:128–33.

13. Aytac A, Yurdakul Y, Ikizler C, Olga R, Saylam A.Pulmonary hydatid disease: report of 100 patients. Ann Thorac Surg 1977; 23: 145–51.

14. Gottstein B, Reichen J. Hydatid lung disease (echinococcosis/hydatidosis). Clin Chest Med 2002; 23: 397–408.

15. Qian ZX. Thoracic hydatid cysts: a report of 842 cases treated over a thirty-year period. Ann Thorac Surg 1988; 46:342–346.

16. Filice C, Brunetti E. Use of PAIR in human cystic echinococcosis. Acta Trop 1997; 64: 95–107.

17. Akhan O, Ozmen MN, Dincer A, Gocmen A, Kalyoncu F.Percutaneous treatment of pulmonary hydatid cysts. Cardiovasc Intervent Radiol 1994; 17: 271–75.

18. Dhaliwal RS, Kalkat MS. One-stage surgical procedure for bilateral lung and hydatid cysts. Ann Thorac Surg 1997;64:338-341.

19. Cetin G, Dogan R, Yuksel M. Surgical treatment of bilateral hydatid disease of the lung via median sternotomy: experience in 60 consecutive patients. Thorac Cardiovasc Surg 1988;36:114-117.

20. Petrov DB, Terzinacheva PP, Djambazov VI. Surgical treatment of bilateral hydatid disease of the lung. Eur J Cardiothorac Surg 2001;19:918–923.

21. Qiu J, Schantz P, Wang Q, He J, Chen X, Liu F. Ozfendazole treatment for experimental alveolar echinococcosis in mice. J Pract Parasit Dis 1999;7:116–19.