

Pulmonary hydatid cyst and successful renal transplantation

Manoucher Aghajanzadeh¹,MD., Ali Monfared²,MD., Gholam-Reza Mokhtari³,MD.,
Masood khosravi⁴, Mohamad K Lebody⁵,MD., Farshid Poorreza⁶,MD.,
Reza Orangpour⁷,MD.

Department of Thoracic Surgery, Guilan University of Medical Sciences Rasht, Iran.

Abstract

Hydatid cysts are endemic hooknoses in Iran. It may involve various organs of body. Liver is involved in 80% and lung in 10- 15%of cases. 25% of pulmonary hydatid cyst is bilateral. Before any transplantations eradication of infection is necessary. In a 26-year old man with renal failure referred for kidney transplantation. On initial preoperative evaluation of this patient hydatid cyst lesion was found in two lungs, left side operated, and right side left in place. Under treatment with albendazole kidney transplantation was performed. After two-year of follow-up, patient was in good condition and the size of right cyst did not increased.

Keywords: pulmonary hydatid cyst, kidney transplantation, immunosuppression, albendazole.

Introduction

Cystic hydatidosis is a zoonosis and a significant public health problem in different areas such as the Middle East [1,2]. Renal transplantation is also not an uncommon procedure now days. Infection is a major complication of renal transplantation. Thus, eradication of any active infection before transplantation is necessary. It is a troublesome situation when a renal failure patient needing transplant surgery suffers from Echinococcus infestation or a hydatid cyst.

The main treatment of pulmonary hydatid cyst is surgical extraction of cyst and, in a few situations; chemotherapy is indicated too [2-7]. Minimally invasive surgery as laparoscopy and percutaneous drainage recommended for liver hydatid cyst [8-11]. Immunosuppressant drugs may allow proliferation of metacestode rem-

nants or proliferation of previous in apparent metastases [1,12-15]. As far as we know, renal transplantation has not been performed in a patient with pulmonary hydatidosis concomitantly. Our extensive literature review could not find renal transplantation in-patient suffering from Hydatid disease too. Almost all studies regarding transplantation and hydatid cyst have been performed on liver transplantation [16-19].

Case report

A young 26-year old man, who was a shepherd admitted in the hospital for chronic renal failure management and kidney transplantation. On initial preoperative evaluation of this patient cystic lesion was found in two lungs, the etiology of chronic renal failure was not known. Results of routine Physical examination and vital signs were normal.

1. **Corresponding author**, Assistant Professor of Thoracic Surgery, Guilan University of Medical Sciences. No. 37, 100th Ave. Golsar Rasht, Iran. Tel: +98131 7722389, email: Maghajanzadeh2003@yahoo.com.

2&4&5. Department of Nephrology, Guilan University of Medical Sciences Rasht, Iran.

3&6. Transplantation and Urology, Guilan University of Medical Sciences Rasht, Iran.

7. General Practitioner.

Chest radiography and CT scan showed a large cyst in left lung (8-4cm) and a small cyst in right side (2-1cm) (Fig. 1). All serology tests for hydatid cyst were negative.

There was not any other cystic mass else where in the body. By poster-lateral thoracotomy surgical extraction of left lung cyst was performed, and then, due to general condition of patient, under treatment with albendazole, renal transplantation was performed from a unrelated donor. The immunosuppressant drug regimen was Scandium, Cellcept and Corticosteroids. Duration of chemotherapy with albendazole (400 mg twice a day) was three course of 28 day with 14 day interval for every course.

During one year follow up after transplantation, The chest radiography and CT-scan did not show any enlargement in cyst size or new cyst formation (Fig. 2).

CT-scan follow up after 2 year of surgery showed no change in size (Fig. 3), and transplant patient was in good condition after three year follow up.

Discussion

Humans are incidental hosts and do not play a role in the biologic cycle. Adult tapeworm inhabits the small intestine of the definitive hosts. The eggs expelled in the feces are infective for susceptible hosts and humans. Then oncospheres hatch from the eggs and penetrate the intestinal mucosa, enter the blood or lymphatic system and Migrate to the visceral organs [1,2].

Both humeral and cellular immune responses are developed against the organism, which results in host protection following the primary infection [1,12-15].

Immune responses occur initially against the oncospheres that penetrate the gastrointestinal mucosa and subsequently against the metacystode [1, 12-15].

Studies suggested that Th1 cell activation is crucial for protective immunity, whereas Th2 cell activation is associated with progressive hydatid disease [1, 12].

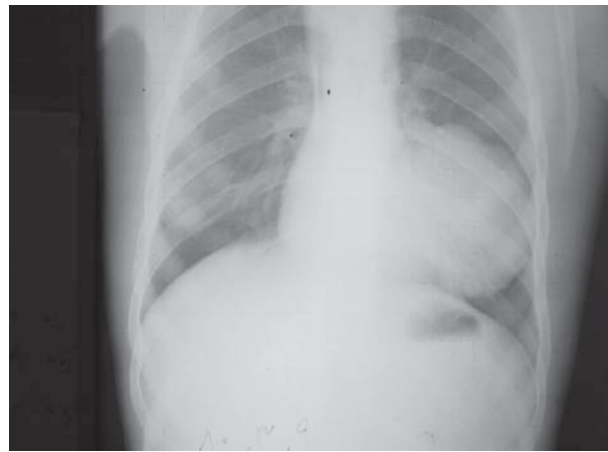


Fig 1: PA CXR of the patient with huge hydatid cyst in left lung and small cyst in right lung.

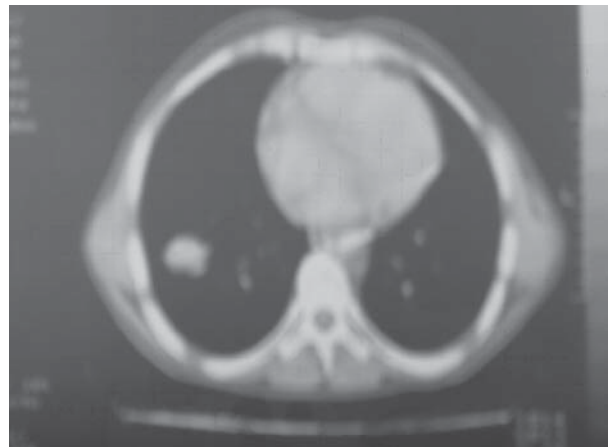


Fig 2. CT scans of the patient one year after transplantation.

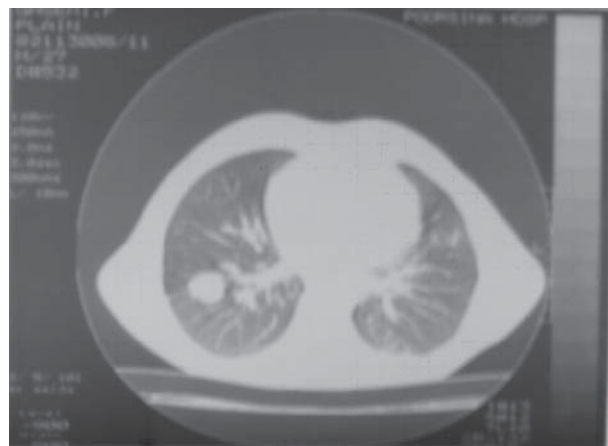


Fig 3. CT scans of the patient two years after transplantation.

With this patient we had two problems: whether the masses of lungs were hydatid cyst or other cystic lesion and if hydatid cyst was alive or dead.

To answer the first question, we reviewed the method of dimaging as an appropriate tool for diagnosis of hydatid cysts. Plain film showed rounded masses of uniform density. The MRI, CT, and ultrasound reveal well-defined cyst with thick or thin wall.

The most pathognomic finding is daughter cyst or laminated membranes and septation in Hydatid Cyst cavity. Presence of daughter cyst, eggshell or mural calcification on CT is indicative of a hydatid infection and helps to distinguish the cyst from carcinoma, bacterial or amebic abscesses and hemangioma [21- 23].

Diagnostic aspiration is not usually recommended in hydatid cyst of lung because of the risk of dissemination and anaphylaxis [2]. But in hydatid cyst of liver percutaneous drainage recommended for diagnosis and treatment [9- 11]. PAIR is a procedure for liver hydatid cyst, and involves several steps. Percutaneous puncture of cysts is performed under ultrasound or CT guidance followed by aspiration of substantial amounts of cyst fluid and injection of a protoscolicidal agent into the cyst cavity (usually hypertonic saline or ethanol)[10]. Laparoscopic treatment of liver echinococcosis has become increasingly popular [8]. Thoracoscopy and PAIR are not recommended for pulmonary hydatid cyst [2].

Detection of antibody to specific echinococcus antigen by immunoblotting has the highest degree of specificity [12- 15].

The lung hydatid cyst in our patient was diagnosed by the CXR and CT-scan, and posterolateral thoracotomy surgical extraction of the left lung cyst was performed.

The imaging criteria of inactivated hydatid cyst included [2, 20- 23].

-No increase in the size of cyst (cysts size typically increase 1-5cm / year).

-Total calcification of the cyst wall (egg shell

appearance)

-Collapsing, flattening, elliptical cyst correspond to low pressure within the cyst

-Detachment of the germinal layer from the cyst wall (water lily sign)

-Coarse echo within the cyst.

Before renal transplantation, it was confirmed that the cyst was inactive by serial CT scan and plain film that showed no increase in size, eliptyform, and calcification.

Responsiveness to pharmacologic therapy should have been established by serology but it was unavailable [12-15]. Almost all studies in transplantation and hydatid cyst have been performed on liver transplantation. Liver transplant is the only option of treatment for alveolar echinococcus, in patients with advanced liver disease due to alveolar echinococcus and because surgery and chemotherapy cannot eradicate the alveolar echinococcus [16-19].

The results of liver transplantation for alveolar Echinococcus on a series of 47 European patients during 1985- 2002 showed that 5 year-survival was 71% and 5 year- survival without recurrence was 58%.

The nine early deaths were involved alveolar echinococcus patients with a long history of symptoms and progressive liver disease such as cholangitis and biliary cirrhosis. Five late deaths were directly related to ongoing Alveolar Echinococcus located in the brain of 3 cases that were not investigated before liver transplantation. [16]. Careful evaluation of possible distant metastases should be done prior to any transplantation. If any active cyst is present, transplantation should be cancelled.

Conclusion

After transplantation in Hydatid Cyst patients the possibility of ongoing echinococcus must be permanently kept in mind that this could be reduced by lightening the immunosuppressant and administrating albendazol before and after transplantation and follow-up of the

patient with specific circulatory markers for hydatid cyst. Chronic stimulation of the host by cyst fluid antigen increased specific IgG 4 production, which might acted as blocking antibody. The less well-defined mechanisms could possibly be presence of parasite-derived-modulating substances such as an anti-complement factor.

Acknowledgement

Thank Gilda Aghajanzadeh for helping us to prepare this manuscript.

References

1. Zhang J L, Donald P. McManus: concepts in immunology and diagnosis of hydatid disease. *Clinical Microbiology Reviews*, January 2003, Vol. 16, No. 1. pp. 18-36
2. Nickolaos NH, Homeros AA, Panagiotis NS. Hydatid Disease of the Lung. *General Thoracic Surgery*. Thomas W. Shields. Lippincott Williams & Wilkins, Sixth ed; 2004. pp.1298-1299.
3. Burgos R, Varela A, Castedo F, Roda J, Montero CG. Pulmonary hydatidosis: surgical treatment and follow-up of 240 cases. *Eur J Cardiothorac Surg*.1999 Dec;16(6): 628-34.
4. Todorov T, Vutova K, Donev A, Katzarov K. The types and timing of the degeneration changes seen in the cysts during and after benzimidazole treatment of cystic echinococcosis. *Ann Trop Med Parasitol* 2005 Oct; 99(7):649-59
5. Dgru D, Kiper N, Ozcelik U, Yalcin E, Gocmen A. Medical treatment of pulmonary hydatid disease: for which child? *Parasitol Int* 2005 Jun; 54(2):135-8.
6. Keraidas D, Mavridis G, Soutis M, Passalidis A. Medical treatment of pulmonary hydatosis: complication and surgical management. *Pediatr Surg Int* 2004 Jan;19(12): 774-6
7. Keshmiri M, Baharvahdat H, Fattahi SH, Davachi B, Dabiri RH, Baradaran H. A placebo controlled study of albendazol in the treatment of pulmonary echinococcosis. *Eur Respir J* 1999 Sep;14(3): 503-7.
8. Georgescu SO, Lazescu D, Duebi L, Crumpei F, Stratan I. Minimally invasive treatment of pulmonary and pleural hydatid cyst. *Rev Med Chir Soc Med Nat Iasi*

2003 Oct-Dec;107(4):893-902 [Romanian].

9. Biswas A, Handa R, Aggarwal P, Wig N, Wali JP. Percutaneous drainage of hydatid cyst an alternative to surgery. *J Assoc Physician India*1998 Jun; 46(6):564-5.
10. Nasser Moghaddam S, Abrishami A, Malekzadeh R. Percutaneous needle aspiration, injection, and reaspiration with or without benzimidazole coverage for uncomplicated hepatic hydatid cysts. *Cochrane Database Syst Rev* 2006; CD003623.
11. lama H, Abdel-Wahab M, Strickland GT. Diagnosis and treatment of hepatic hydatid cysts with the aid of echo-guided percutaneous cyst puncture. *Clin Infect Dis* 1995; 21:1372.
12. Baz A, Richieri A, Puglia A, Nieto A, Dematteis S. Antibody response in CD4-depleted mice after immunization or during early infection with *Echinococcus granulosus*. *ParasiteImmunol*1999; 21:141-148
13. Aceti A, et al. IgG subclasses in human hydatid disease: prominence of the IgG4 response. *Int. Arch Allergy Immunol*1993; 102:347-351
14. Heath DD, Lawrence SB. Antigenic polypeptides of *Echinococcus granulosus* oncospheres and definition of protective molecules. *Parasite Immunol* 1996. 18:347-357.
15. Chemtai A. K., Okelo G.B, and Kyobe J. Application of immuno-Electrophoresis (IEP5) test in the diagnosis of human hydatid disease in Kenya. *East Afr Med J* 1981; 58:583-586
16. Bresson-Hadin S, Koch S, miguet JP, Gillet M, Mantion GA, Heyd B, Vuitton DA. Indication and results of liver transplantation for *Echinococcus alveolaris* infection: an overview. *Langenbecks Archives of Surgery*.2003; Vol 388, Number 4 Page: 231-238
17. Delines for treatment of cystic and alveolar echinococcosis in humans. WHO Informal Working Group on Echinococcosis. *Bull World Health Organ* 1996; 74:231.
18. Gottstein B. Epidemiology and systematics of cystic and alveolar hydatid disease. *Chirurg* 2000 Jan; 71(1):1-8.
19. Renner, EC, Bachmann, LM, et al. Evaluation of treatment and long-term follow-up in patients with hepatic alveolar echinococcosis. *Br J Surg* 2005; 92:1110.
20. Babba H, Messedi A, Mahoudi S, Zribi M, Grillot R, Ambriose-Thomas P, Beyrouiti I, and Sahnoun Y. Diagnosis of human hydatidosis: comparison between imaging and six serologic techniques. *Am. J. 4-Trop. Med. Hyg.* 1994; 50:64-68
21. Kervancioglu, R, Bayram, M, Elbeyli, L. CT findings in pulmonary hydatid disease. *Acta Radiol* 1999; 40:510.
22. Marani, SA, Canossi, GC, Nicoli, FA. Hydatid disease: MR imaging study. *Radiology* 1990; 175:701.
23. Von Sinner WN, Rifai A, Te Strake L, Sieck J.

Magnetic resonance imaging of thoracic hydatid disease. Correlation with clinical findings, radiography, ultrasonography, CT and pathology. Acta Radiol 1990; 31:59.