

An Unusual Presentation of Hydatid Cysts in a Four Year Old Child

Fatima Ismail¹, Ayesha Mansoor²

Abstract

We report case of 4 years old vaccinated girl weighing 13 kg who was admitted in our hospital with complaints of fever & cough off and on for last 1 year. This was accompanied with difficulty in breathing for past 1 week only. Her Chest x-rays demonstrated left sided lobar consolidation. She responded to antibiotics but complete resolution of the consolidation was not observed. On repeat chest X ray on day 7, the radiological findings were same, following which an ultrasound chest was done which shows cystic lesion on left side of the chest and another cyst in upper abdomen, we confirmed this with computerized tomography (CT) chest and CT abdomen. Serological tests for hydatid cyst (indirect haemagglutination) were positive. A provisional diagnosis of hydatid cyst was made and the child was put on albendazole. A surgical consult was also taken. After 6 months of albendazole off and on, due to non-compliance, surgery was done. Cyst of liver of removed successfully and albendazole was continued. Child responded and repeat radiological survey showed a marked decrease in the size of the cyst and at present the child is clinically well and now going regularly to school.

We are reporting this case as it presented in an unusual manner and the child had been seen by a number of physicians, and given various antibiotics, with no response and finally the parents brought the child to our hospital. The hydatid cyst in this young girl was missed out because it was not considered in the differential diagnosis of persistent breathlessness.

Keywords: Hydatid cyst, paediatric, consolidation, albendazole. (ASH & KMDC 19(2):97;2014).

Introduction

Echinococcus granulosus is a cestode parasite (tapeworm) that causes hydatid infection. These cestodes have a worldwide distribution but the prevalence is higher in developing countries^{1,2}. Infection rate is as low as 1 per 1000 in North America and as high as 10% in the third world. This disease is endemic in cattle and sheep rearing regions of the world. The close association of people with sheep and dogs and the unavailability of clean potable water supplies in Pakistan make it a region endemic to the disease³. It is a parasitic in-

fection caused by the Cestode tapeworm *Echinococcus granulosus*. The dogs are the definitive host and the adult worms are found in their small intestine. Humans get infected either by contact with the definitive host or by consuming vegetables and water contaminated with the hydatid ova⁴. Most common symptoms are cough, dyspnoea, fever, chest pain and haemoptysis. However, these are not specific findings for hydatid disease⁵.

Theoretically, echinococcosis can involve any organ. The liver is the most common organ involved, followed by the lungs. These two organs account for 90% of cases of echinococcosis and other viscera including kidney, spleen, heart and brain account for other 10%⁶⁻⁸.

Diagnosis can be made by Complete Blood Count (CBC) which will detect eosinophilia in 30% of patients, plain abdominal x-ray which may show

¹⁻²Department of Paediatrics Unit II
KMDC & Abbasi Shaheed Hospital Karachi

Correspondence: Dr. Fatima Ismail
House Officer, Paediatric Unit II,
Abbasi Shaheed Hospital and
Karachi Medical & Dental College
Cell: 0303-9088503
Email:fatima_028@yahoo.com

calcification in cyst wall while the cyst can also be imaged with ultrasound or CT. It can be confirmed by indirect haemagglutinin assay^{9,10}.

We report a case evaluated for non resolving pneumonia in which diagnosis was made by serology and CT scan.

Case Report

Four year old girl weighing 13 kg vaccinated presented with fever & cough off and on for last one year, with difficulty in breathing for last one week. She took multiple oral antibiotics which temporarily relieved these symptoms. She visited many doctors and private hospital but was not adequately investigated or managed. For the last one week she had developed difficulty in breathing for which they came to Abbasi Shaheed Hospital and child was admitted. During course of illness her appetite was reduced, there was a history of weight loss, but no history of similar illness in the family. She had no other significant history. There was no history which was suggestive of pulmonary tuberculosis. Physical examination revealed a patient with respiratory distress, Blood Pressure (BP) of 80/60 mm Hg, heart rate of 160 beats per minute and temperature of 101°F. Her chest examination revealed diminished movements over the left side, intercostal recessions were present. On percussion, there was dull note on left side of chest. On auscultation, air entry decreased on a left side of chest, increased vocal resonance at left lower chest and bilateral crepitations were present. The abdomen was slight tender in epigastrium with palpable liver of 6 cm and total span of 12 cm with regular margins and smooth surface and slightly tender. Her cardiovascular examination and neurological examination was unremarkable. Her CBC shows Hb 9.4 mg/dl MCV 78 fl, MCHC 35 pg, WBC 14000 uL⁻¹, Lymphocytes 38.2%, Granulocytes 56.7%, 3% Eosinophils. Haematocrit 28, Platelet 230,000 and ESR 25 mm. Mountoux Test was negative.

Her chest radiograph showed showing consolidation in the upper and middle lobe of the left lung Table/Fig.1. Initially, we made a diagnosis of left

sided lobar pneumonia, and she was kept on IV antibiotics, patient became afebrile and her general condition improved. On repeat chest X ray on day 7, the radiological findings were same, then we did Ultrasound chest which shows a large cyst seen in left upper and middle zone measuring 7 into 6 cm and another cyst in upper abdomen 7.4 into 6.4 cm. Computed Tomography (CT) of chest and abdomen confirmed the same (Fig. 2, Fig. 3). CT chest and abdomen showed an ill defined oval shape cystic lesion with enhancing walls seen in left upper lobe, it measures about 7 into 6 cm, no enhancing solid component and septa seen. Air space shadowing with air bronchogram is seen in right middle lobe suggestive of consolidation. There is well defined round hypodense area measuring about 6.5 into 5.5 cm seen in left lobe of liver minimal rim enhancement seen. No enhancing solid component septa seen. There is no evidence of intra and extra hepatic duct biliary dilatation.

The diagnosis was made of hydatid cyst in lung and liver and then confirmed by Echinococcus antibody titre of 1:512 which was positive.

The patient was given albendazole and was referred for surgical opinion to NICH. After 6 months of albendazole off and on, due to non-compliance, surgery was done. Cyst of liver was removed successfully. Lung remained as such and received albendazole was continued for one year. The child was fine after the treatment and is now active, going to school on a regular basis. Parents refused any further investigations after one year such as X-ray or CT scan.

Discussion

Hydatid cysts most often develop in the liver. However when embryos pass through this first filter, the second most frequent location is the lung. Hydatid cysts can occur anywhere in the body. Hydatid cysts are typical, involving one lobe in 72% of cases, usually at the lung base.

Coughing, chest pain, and breathlessness are common presenting features of the pulmonary disease¹¹. History of animal contact (especially dogs)

and living in a sheep-raising or cattle-raising rural area is generally present. Majority of the cases of hydatid disease are seen coming from rural areas or people settled in urban centers after spending life in villages but in our case the patient was from urban city like Karachi and without any history of contact with sheep or dog.

Most of the people acquire the disease during their childhood but do not present the clinical signs and symptoms until late adulthood¹². The reported cases in pediatrics have an age group between 6 months to 9 years and the average age is 5 years in Pakistan¹³⁻¹⁶. The case which we have described here is of younger age than average and presented with rapid course of symptoms than usual.

A plain radiograph of the region involved may show an unruptured cyst appearing as a spherical, well-circumscribed, homogenous opacity, particularly pronounced if calcified. Blood eosinophilia is reported to occur in 20% to 34% of patients¹⁷.

Ultrasonography (US) and CT have been reported to be the main diagnostic tools, with 85% and 100% sensitivity, respectively¹⁸. Although US is a reliable method for detecting echinococcal cysts, CT scans can define unique characteristics of hydatid cysts while also revealing additional small unsuspected lesions. Every patient who has Hydatid cysts of the lung should be investigated for associated cysts in the liver or in other abdominal organs by chest CT and abdominal ultrasonography, as were performed for the case which is under discussion. Pre-operative diagnosis of Hydatid cysts can be made ultrasonically and confirmed by a CT scan. The serological tests are complementary to clinical and radiologic findings and can also be used in the follow-up of patients after surgical resection¹⁹. The serological tests including Casoni intradermal skin test, Weinberg complement fixation (CF) test, indirect hemagglutination (IHA) test, ELISA, and western blot (WB) are the frequently used laboratory tests for diagnosis of Hydatid disease, with the reported sensitivity of 96.7%, 87.1%, and 100%, for IHA, ELISA, and WB, respectively²⁰.

Medical treatment with an antihelminthic agent benzimidazole compound, either mebendazole or albendazole, is usually administered for the established Hydatid disease. Albendazole is known to be more effective than mebendazole²¹.

A standard dose of albendazole is 10 to 15mg/kg/d (taken twice daily). Owing to its hepatotoxicity, a one week to two weeks interval should be given between three weeks and four weeks cycles and treatment may last for 3 to 6 months. In some cases, praziquantel can be added to albendazole.

The treatment of Hydatid cysts is principally surgical resection especially in the cases of cysts in lung and abdomen causing compression as it was in our case too. But better forms of chemotherapy and methods like Puncture, aspiration, injection and reaspiration (PAIR) technique are also other means of management²². However, pre- and post-operative one month courses of Albendazole and 2 weeks of Praziquantel should be considered in order to sterilize the cyst, decrease the chance of anaphylaxis, decrease the tension in the cyst wall (thus reducing the risk of spillage during surgery) and to reduce the recurrence rate post-operatively.

We conclude that *Echinococcus granulosus* can affect any organ in the body and a high suspicion of this disease is justified in any case, resolving pneumonia especially in endemic regions. Moreover, medical treatment should precede and follow the surgical intervention. Complicated pulmonary Hydatid cysts imitate several pleura and pulmonary diseases such as non resolving pneumonia, tuberculosis, abscess and tumor.

The diagnosis of the Hydatid cyst was made mainly with the help of imaging methods and serology. Although the diagnosis is confirmed by cytology or histology, imaging modalities such as ultrasonography, CT and MRI are more sensitive than serologic tests and in these cases; the involvement of other organs should be investigated.

Fig. 1 : Chest radiograph showing consolidation in the upper and middle lobe of the left lung

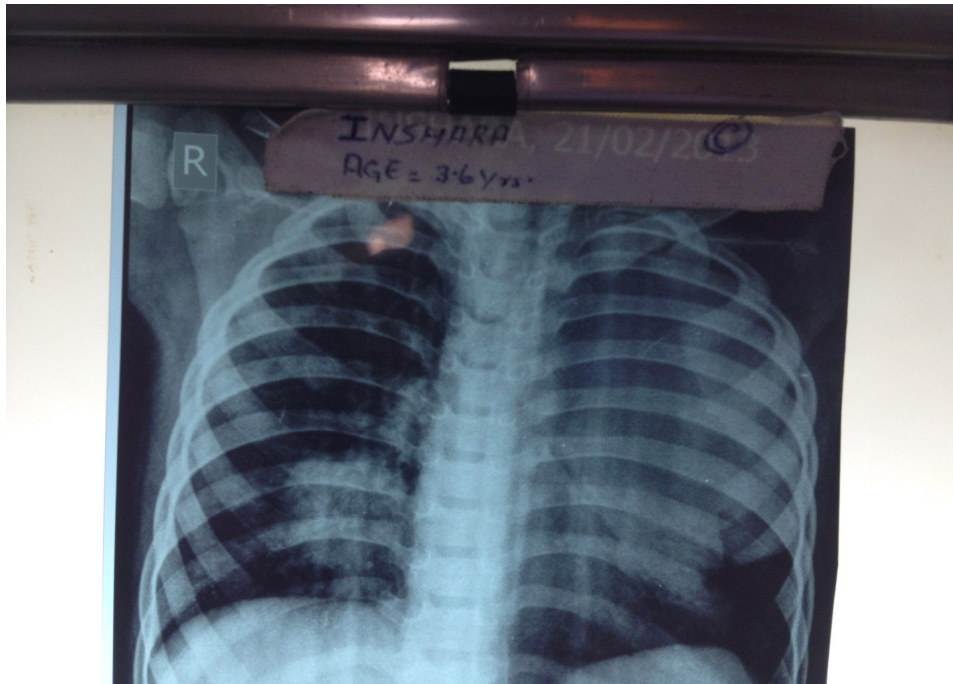
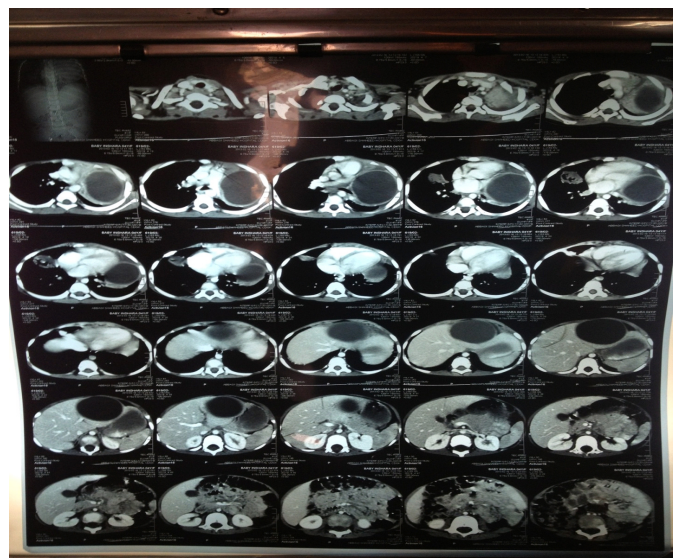
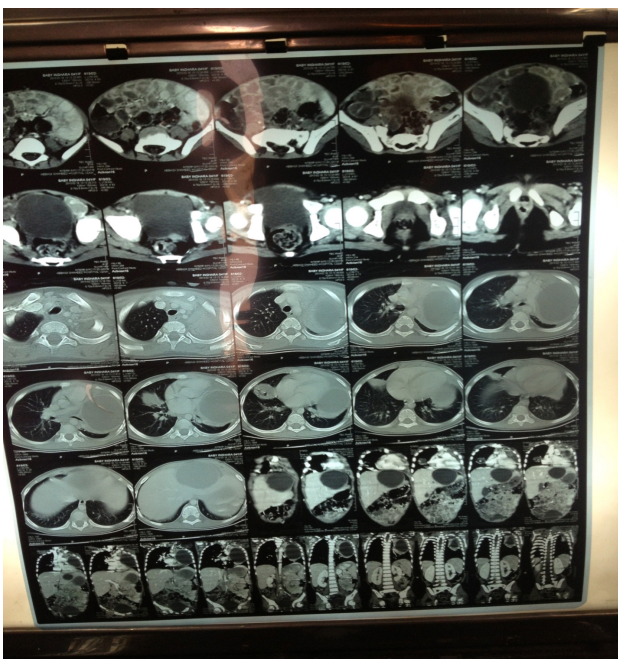


Fig. 2 : CT chest and abdomen showed an ill defined oval shape cystic lesion with enhancing walls seen in left upper lobe 7 x 6 cm. Air space shadowing with air bronchogram is seen in right middle lobe suggestive of consolidation. There is well defined round hypodense area measuring about 6.5 x 5.5 cm seen in left lobe of liver minimal rim enhancement seen.



Conclusion

Hydatid cyst has varied presentations. Hydatid cyst is prevalent not only in rural areas but also reported in urban areas. Multiple cysts can be present in a patient in different locations. Imaging techniques are main diagnostic modalities IHA titer varies and negative value does not rule out hydatid cyst. Surgery is still found to be a best modality

This case contributes to the literature in an aspect that hydatid cyst should be kept in differential diagnosis of non resolving pneumonias. Familiarity with atypical manifestations of hydatid disease may be helpful in making a prompt, accurate diagnosis.

References

- Pedrosa I, Saiz A, Arrazola J, Ferreiros J, Pedrosa CS. Hydatid disease: Radiologic and pathologic features and complications. *Radiographics* 2000;20:795-817.
- Ismail MT. Hydatid Cysts in Children. *Annals of Pediatric Surgery* 2010;6: 98-104.
- Hussain A, Maqbool A, Hussain S, Athar M, Shakoor A, Amin MK. Studies on prevalence and organ specificity of hydatidosis in ruminants slaughtered at Karachi and Faisalabad abattoir, Pakistan. *Indian J Dairy Sci* 1992;45:454-6.
- Polat P, Kantarci M, Alper F, Suma S, Koruyucu MB, Okur A. Hydatid disease from head to toe. *Radiographics* 2003;23:475-94.
- Morar R, Feldman C. Pulmonary echinococcosis. *Eur Respir J* 2003;21:1069-77.
- Kiresi DA, Karabacakoglu A, Odev K, Karakose S. Uncommon locations of hydatid cysts. *Acta Radiol* 2003;44:622-36.
- Engin G, Acunas B, Rozanes I, Acunas G. Hydatid disease with unusual localization. *Eur Radiol* 2000;10:1904-12.
- Husen YA, Nadeem N, Aslam F, Bhalla I. Primary splenic hydatid cyst: a case report with characteristic imaging appearance. *J Pak Med Assoc* 2005;55:219-21.
- Ayles HN, Corbett EL, Taylor I, Cowie AG, Bligh J, Walmsley K. A combined medical and surgical approach to hydatid disease: 12 years' experience at the Hospital for Tropical Diseases, London. *Ann R Coll Surg Eng* 2002;84:100-5.
- Filippou D, Tselepis D, Filippou G, Papadopoulos V. Advances in liver echinococcosis: diagnosis and treatment. *Clin Gastroenterol Hepatol* 2007;5:152-9.
- Tekinbas C, Turedi S, Gunduz A, Erol MM. Hydatid cyst disease of the lung as an unusual cause of massive hemoptysis: a case report. *J Med Case Rep* 2009;3:21.
- Todorov T, Boeva V. Echinococcus in children and adolescents in Bulgaria: a comparative study. *Ann Trop Med Parasitol* 2000;94:135-44.
- Ali R, Sajjad R, Sina S. Surgical treatment of pulmonary Hydatid cyst in children. *Tanaffos* 2009;8:56-61.
- Jain P, Sanghvi B, Shah H, Parelkar SV, Borwankar SS. Thoracoscopic excision of mediastinal cysts in children. *J Minim Access Surg* 2007;3:123-6.
- Halezeroglu S1, Celik M, Uysal A, Senol C, Keles M, Arman B. Giant hydatid cysts of the lung. *J Thorac Cardiovasc Surg* 1997;113:712-7.
- Talaiezadeh AH, Maraghi SH. Hydatid disease in children: A different pattern than adults. *Pakistan J Med Sci* 2006;22:329-32.
- Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. *Clin Microbiol Rev* 2004;17:107-35.
- Turgut AT, Altin L, Topcu S, Kilicoglu B, Aliinok T, Kaptanoglu E, et al. Unusual imaging characteristics of complicated hydatid disease. *Eur J Radiol* 2007;63:84-93.
- Gunay K, Taviloglu K, Berber E, Ertekin C. Traumatic rupture of hydatid cysts: a 12-year experience from an endemic region. *J Trauma* 1999;46:164-7.
- Hernandez-González A, Muro A, Barrera I, Ramos G, Orduna A, Siles-Lucas M. Usefulness of four different echinococcus granulosus recombinant antigens for serodiagnosis of unilocular hydatid disease (UHD) and postsurgical follow-up of patients treated for UHD. *Clin Vaccine Immunol* 2008;15:147-53.
- Taylor DH, Morris DL. In vitro culture of *Echinococcus multilocularis*: protoscolicidal action of praziquantel and albendazole sulphoxide. *Trans R Soc Trop Med Hyg* 1988;82:265-7.
- Tan A, Yakut M, Kaymakcioglu N, Ozerhan IH, Cetiner S, Akdeniz A. The results of surgical treatment and percutaneous drainage of hepatic hydatid disease. *Int surg* 1998;83:314-6.