Ruptured pulmonary hydatid cyst - masquerading as tuberculous pleural effusion in a child

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Abstract

We report a case of intrapleural rupture of hydatid cyst of the lung in a 12 year old female child who presented with a persistent cough since 3 years. She was misdiagnosed as tuberculous pleural effusion, which on contrast enhanced computed tomography thorax was diagnosed as ruptured hydatid cyst. She was treated conservatively and referred for further surgical correction. Following the surgical correction, the patient condition improved dramatically.

Key words: Hydatid cyst, Lung, Intrapleural rupture

ydatid disease is caused by infection with the larval stage of the dog tapeworm Echinococcus granulosis. In children, pulmonary disease is common and peaks between 5 and 15 years of age. 30% of these cysts may rupture producing pleural effusions, bronchial seeding, and occasionally acute anaphylaxis. Intrapleural rupture is relatively rare presentation observed in only 1.5-6% of cases [1]. The diagnosis becomes very difficult if there are no clinical symptoms or signs suggestive of hydatid disease and considering the differential diagnosis of pleural effusion (tuberculosis, carcinomatosis, infection etc.). We report such a case of pulmonary hydatid cyst with intrapleural rupture, which was initially misdiagnosed as tuberculous pleural effusion.

CASE REPORT

A 12-year-old female child presented with a history of persistent cough and low grade fever for past 3 years, which was associated with copious amount of watery and sometimes whitish sputum production. It started with an episode of aspiration of milk during breakfast. Following which the cough persisted, and she was treated by multiple physicians with different antibiotics over the period of 3 years with no improvement. She also received antitubercular drugs for 9 months in view of incomplete response to antibiotics and chest X-ray, which showed homogenous opacity involving the right side of the chest. However, other investigations to support the diagnosis of pulmonary tuberculosis were normal. The patient remained asymptomatic (i.e. without any cough or fever) for about 2 months, following which the symptoms

reappeared. Now, she was treated as tubercular relapse and anti-tuberculosis treatment was continued for another 9 months along with oral dexamethasone. As there was no improvement, the patient was also started on herbal medicines in November 2013.

However, she still got no relief and developed blood stained sputum production 6 months back for which she was brought to our hospital. She also had difficulty in breathing even at rest for last 3 days. There was no history of fever, chest pain, night sweats, loss of appetite or loss of weight. She belonged to a rural area, and there was no history of contact with tuberculosis and she was immunized as per the age. There was a history of rearing pet dog in the family, which was earlier a stray dog and is not immunized.

On examination, the patient was afebrile, conscious, alert, moderately built and nourished with mild pallor and no lymphadenopathy. Vital signs were pulse rate: 112/min, respiratory rate: 44/min, blood pressure: 112/54 mm-Hg. Respiratory system examination revealed diminished chest movement, stony dullness on percussion, diminished breath sounds on auscultation over the right side of the chest. Trachea was in the midline. On other system examination, no abnormality was detected.

We managed her conservatively with oxygen, intravenous antibiotics, intravenous fluids and antipyretic. Her plain X-ray chest revealed a massive homogenous opacity involving whole right side of the chest sparing some part of the upper

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lobe of the lung. There were no mediastinal shift and no shadow of a collapsed lung. Blood investigations showed haemoglobin - 11.8 g/dl, total leukocyte count: 7,950/mm³, differential count: Neutrophils - 79%, lymphocytes - 18%, monocytes - 3%, and platelets - 3.77 lakhs/mm³. Liver function tests and renal function tests were within normal limit. Her sputum analysis for AFB was negative. As we suspected it to be something other than effusion, we proceeded with contrast enhanced computed tomography (CT) - thorax which revealed a large hydatid cyst of approximate size $14 \text{ cm} \times 12 \text{ cm} \times 9 \text{ cm}$ with air fluid level and floating membrane inside involving right whole lung sparing the apical region suggestive of intrapleural rupture. Another hydatid cyst of size $4.7 \text{ cm} \times 4.6 \text{ cm}$ in left lower lobe was also found.

Patient was referred for surgical management and was treated by pleural decortication and evacuation of hydatid membrane. Postoperatively, oral albendazole was added at a dose of 400 mg twice a day, which was to be continued for 6 months. The patient recovered dramatically within 2 weeks of the treatment and there was no cough or respiratory problems. The parents are counseled regarding exposure to dogs, cattles and maintenance of proper hygiene practices and chest physiotherapy.

DISCUSSION

Echinococcosis (hydatid disease or hydatidosis) is the most widely spread serious human cestode infection in the world [2]. The cysts can be found in the liver, spleen and lungs. The ingested ova burrow through intestinal mucosa and travel to the liver through mesenteric veins. A few ova bypass liver and are trapped in the lung. Most patients who present with pulmonary disease are children [3]. These cysts should be removed because 30% of these lesions may eventually rupture [4], producing pleural effusions [5], bronchial seeding and occasionally acute anaphylaxis [6]. Intrapleural rupture of pulmonary hydatid cysts is relatively rare (1.5-6%) [1]. It is a severe complication with dissemination and high risk of recurrence and overinfection with empyema. It also represents a clinical challenge for diagnosis considering the differential diagnosis of pleural effusion (tuberculosis, carcinomatosis, infection etc.). The rupture is most often primary from a superficial and large lung lesion and sometimes secondary to trauma or accidently after needle aspiration [7].

In acute phase, chest X-ray can show a pneumothorax, hydropneumothorax or hydrothorax. The diagnosis becomes very difficult if there are no clinical symptoms or signs suggestive of hydatid disease. In such cases, a pleural tap may be the only way to prove its presence [1]. In uncomplicated hydatid cysts, radiological diagnosis is relatively easy. CT provides further information in equivocal cases by revealing the fluid density of an intact cyst and the air/fluid density of a ruptured cysts. Simple hydatid cysts have water density on CT and ruptured cysts may

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present with a wide variety of radiological appearances due to different combinations of collapsed membrane, air, and fluid. However, infection of the cyst may increase the attenuation values and produce a solid appearance, which may hamper the correct diagnosis. Such complicated cysts, in the absence of positive history, serological tests and other radiological signs, may simulate a malignant tumor, tuberculosis, abscess or other infected cystic lesions of the lung. The "air bubble sign" has been described in complicated cysts and reported to be an important clue in the differentiation of hydatid cysts from other disease processes [8,9]. Radiological signs occurring as a result of separation of cyst membranes have been described, and include the "crescent," "water-lily," "daughter cysts," "double arch," "ring within a ring," "serpent" or "snake," and "spin" or "whirl" signs [10].



Figure 1: Plain X-ray chest posterior-anterior view, massive homogenous opacity involving whole of the right side of the chest sparing some part of the upper lobe of the lung. There was no mediastinal shift and no shadow of a collapsed lung



Figure 2: Contrast enhanced computed tomography – Chest, a large hydatid cyst of approximate size 14 cm \times 12 cm \times 9 cm with air fluid level and floating membrane inside involving right whole lung sparing the apical region suggestive of intrapleural rupture. Another hydatid cyst of size 4.7 cm \times 4.6 cm in left lower lobe was also found

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Surgery is the only and best form of treatment. After evacuation of fluid in the chest, pleural decortication with extraction of hydatid membrane is performed in order to reduce recurrence and promote full re-expansion of the lung. In cases with pleural effusion following rupture, a temporary chest tube insertion can be done preceding thoracotomy or any other radical resection procedures [1]. Medical treatment with albendazole or mebendazole is used together with surgical treatment to avoid recurrence. It is given pre-operatively to prevent the consequences of possible rupture of the cysts during surgery, and postoperatively as adjuvant therapy for cysts that may have ruptured during the operation [11]. The usual dosage of orally administered albendazole is 10-15 mg/kg/day in two divided doses, or a fixed dose of 400 mg twice a day. For mebendazole the daily dosage is 40-50 mg/kg/day in three divided doses [12].

CONCLUSION

Ruptured pulmonary hydatid cyst masquerading as tuberculous pleural effusion makes this case an interesting one and it teaches us to suspect and diagnose a ruptured hydatid cyst in any unusual and unresolving case of pleural effusion that we encounter in our practice, as this is a benign disease, which can be cured with best results if intervened at an early stage.

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