Case Report

Hydatid lung cysts with hemoptysis mimicking pulmonary tuberculosis

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ABSTRACT

Hydatid cyst of the lung is one of the rare causes of hemoptysis and often diagnosed late. Cystic echinococcosis is the larval cystic stage (echinococcal cysts) of a small taenia-type tapeworm (*Echinococcus granulosus*) that may cause illness in intermediate hosts, generally herbivorous animals and people who are infected accidentally. We present the case of multiple hydatid cysts in a 10-year-old boy presenting with hemoptysis from an endemic area but with no history of contact. Radiological investigations done were inconclusive, and the patient was started on anti-Koch's Therapy (AKT) based on a high clinical suspicion. Surgical intervention was planned after 4 months of therapy due to the non-resolution of symptoms and progression of the disease. Histopathology was suggestive of multiple hydatid cysts occupying the right lung lower lobe. The patient was started on Tab Albendazole and the follow-up period was uneventful. A possibility of hydatid cyst in cases of lung infections should be kept in endemic areas irrespective of the history and radiological features. Complete evaluation, along with tissue diagnosis, is important for establishing a confirmative diagnosis of tuberculosis. Empirical treatment with AKT, in cases of suspicion, should be avoided.

Key words: Hydatid disease, Hydatid lung cyst, Lung cyst

Hydatid cyst of the lung is one of the rare causes of hemoptysis and often diagnosed late. Cystic echinococcosis is the larval cystic stage (echinococcal cysts) of a small taenia-type tapeworm (*Echinococcus granulosus*) that may cause illness in intermediate hosts, generally herbivorous animals and people who are infected accidentally. Pulmonary hydatid cysts typically involve one lobe, usually at the lung base, in 72% of cases. In the pediatric population, males are affected more than females [1-3]. The lungs are the most common site of involvement in children and the second most common in adults [4]. Intact cysts are usually asymptomatic and found as a chance occurrence on chest radiographs. Occasionally, the rupture of the cyst may be the first manifestation of the disease, and this can have catastrophic allergic sequelae [5,6].

We present the case of multiple hydatid cysts in a 10-year-old boy presenting with hemoptysis from an endemic area but with no history of contact. Hydatid lung cyst in the pediatric group can mimic pulmonary tuberculosis, and radiological investigations are the mainstay in diagnosis.

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CASE REPORT

A 10-year-old male child presented to us in December 2017 with chief complaints of cough for 10 days along with hemoptysis for 2 days (two episodes). The child had no history of fever, weight loss, breathlessness, loss of appetite, family history of asthma/tuberculosis, or any history of contact.

On general examination, the patient was conscious, oriented, and responsive to time, place, and person. The patient had a normal gait. There was no pallor and cyanosis. Vitals were stable with a pulse rate of 92/min, blood pressure of 100/70 mmHg, respiratory rate of 18/min, and afebrile.

The chest X-ray was done, which was suggestive of a cavitary lesion in the right lower lobe (Fig. 1a). Induced sputum was sent for acid-fast bacilli (AFB) staining and Cartridge Based Nucleic Acid Amplification Test (CBNAAT) studies, both were reported negative. High-resolution computed tomography (HRCT) chest was done which revealed mass-like consolidation in the right lower lobe with surrounding ground-glass opacity patches, and multifocal tree-in-bud opacities suggestive of infective etiology (Fig. 1b). Repeated samples of induced sputum were sent for

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CBNAAT, and the patient was continued on cough medications. None of the samples were reported positive.

The patient was started on anti-Koch's Therapy (AKT)-category 1 and was discharged. Regular follow-ups were maintained, and the patient showed improvement clinically, hemoptysis stopped, and cough was reduced significantly. After the intensive phase (2 months) of AKT, a repeat chest X-ray was done, which had no resolution radiologically.

After 4 months of AKT (intensive phase of 2 months and continuation phase of 2 months), the patient started having complaints of cough, more during night time, not associated with distress but associated with sticky, scanty, and yellowish sputum. The patient also had hemoptysis, 10–15 ml of fresh blood initially that later increased up to 25–30 mL blood in 24 h. The patient was readmitted and evaluation was done for the cause of reappearance of symptoms.

On investigation, hemoglobin was 9.9 g%, hematocrit was 34.4%, white blood count was 7800/mm^3 (48/42/6/4/0), platelet count was 272,000/mm^3, and erythrocyte sedimentation rate was 22 mm/h. AFB and CBNAAT were negative. Chest X-ray after deterioration suggested progression of pathology (Fig. 2a). Repeat HRCT chest was suggestive of irregular heterogeneous attenuation lesion in the apical region of the right lower lobe with perilesional speculations (Fig. 2b). Follow-up tests revealed the progression of the disease. Fine-needle aspiration cytology (FNAC) of the lesion was done, which was suggestive of inflammatory pathology. FNAC sample was also sent for AFB staining and was reported negative. On correlating FNAC, HRCT, and chest X-ray, the following differentials were considered: Congenital cystic-adenomatoid malformation, pulmonary tuberculosis with bronchiectasis, pulmonary neoplasm, and infected pulmonary cyst.

Surgery opinion was taken; the patient was transferred under the surgery department, and planned for video-assisted thoracoscopic surgery which was converted into the right posterolateral thoracotomy. Intraoperative findings were as follows: Multiple cystic lesions occupying the whole of the right lower lobe, normal right upper and middle lobes, and no adhesions/pus. The right lower lobectomy was done (Fig. 3a), all air leaks checked and repaired. Thoracic toilet was done, hemostasis ascertained, wide bore intercostal drainage (ICD) tube placed. A post-operative X-ray was taken (Fig. 4a) and the sample was sent for histopathology examination.

On macroscopic examination, the specimen was right lung lobe measuring 9.5 cm × 7.5 cm × 5 cm, externally showing multiple sub-pleural cysts ranging from 0.6 cm to 2.5 cm diameter. Culture and sensitivity of small cystic lesions revealed a pearly white cyst wall with a small daughter cyst in the lumen. Separate thin flap like cyst wall pieces measuring 5 cm × 4.5 cm × 1.3 cm showed a glistening surface of gray-white in color (Fig. 3a). Lung on the serial cut section showed a central cystic lesion measuring 4 cm × 3 cm × 2.5 cm with a granular inner surface. On microscopic examination, the cyst wall showed a chitinous lamellated layer (Fig. 3b), and the germinal layer was not distinctly evident.

All these features were suggestive of multiple hydatid cysts occupying the right lung lower lobe. The patient was started on Tab Albendazole. ICD was removed on post-operative day 7 and the patient was discharged after 10 days. The follow-up period was uneventful, air entry improved and there was no recurrence. On follow-up after 4 months, bilateral air entry improved and X-ray was significantly improved (Fig. 4b).

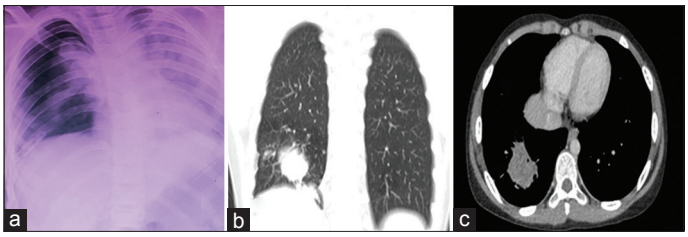

Figure 1: (a) X-ray and (b) high-resolution computed tomography chest suggestive of mass-like consolidation in the right lower lobe lung consolidation

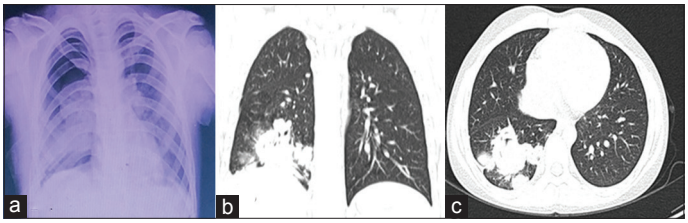

Figure 2: (a) Chest X-ray after deterioration and (b) repeat high-resolution computed tomography chest were suggestive of the irregular lesion in the apical region of the right lower lobe. These findings were suggestive of progression of the disease

DISCUSSION

Respiratory infections are one of the leading causes of infant morbidity and mortality; and in this context, cough is an important initial symptom. Hemoptysis is also an important symptom in

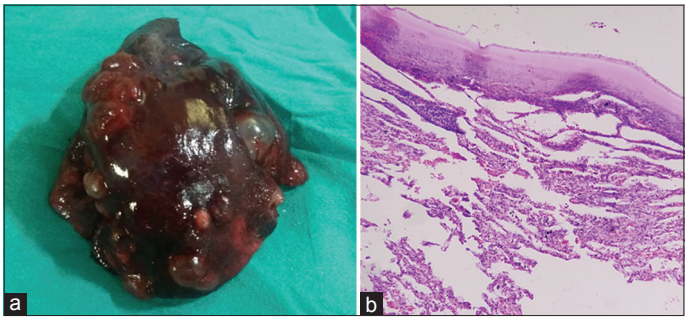

Figure 3: (a) Lobectomy right lung done. Intraoperative picture of the excised specimen showing multiple sub-pleural cysts; (b) histopathological image showing the three layers of the cyst

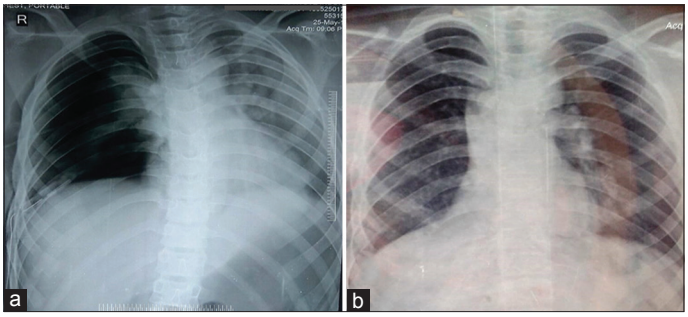

Figure 4: (a) Immediate post-operative X-ray and (b) follow-up X-ray after 4 months

respiratory syndrome associated with tuberculosis [7]. However, a possibility of hydatid cyst in cases of lung infections should be kept in endemic areas irrespective of the history and radiological features.

Hydatid disease represents the larval form of the canine intestinal tapeworm *E. granulosus* [8], which infects humans as accidental intermediate hosts. The adult phase of *E. granulosus* occurs in dogs and other carnivores. The head consists of a double crown of hook-like structures, and the body is formed of three or four rings, the last of which bears the eggs. After being shed in feces, the eggs contaminate fields, irrigated land, and wells. Herbivores such as sheep ingest the eggs, which develop into larvae, or hydatid, in the viscera. The cycle is completed with the ingestion of the infected viscera by carnivores. In accidental cases, humans become aberrant intermediate hosts, contracting the disease from water or food, or by direct contact with dogs. After eggs reach the stomach, embryos released to pass through the intestinal wall, reach the liver, and develop into hydatids. If they advance beyond the liver, they may become lodged in the lung, where they also transform into hydatid. If they overcome the pulmonary obstacle, they may remain in any organ to which they are carried by the bloodstream. There is evidence that embryos can bypass the liver and reach the lung through the lymphatic vessels [9]. Structurally, the cysts consist of a tough outer pericyst that protects a delicate inner endocyst from which brood capsules and daughter cysts develop.

Most intact cysts are seldom symptomatic but may produce chronic cough, dyspnea, pleuritic chest pain, and hemoptysis. Cough, chest pain, and breathlessness are the most common presenting symptoms, while hemoptysis is rare [10]. The mechanism of hemoptysis in pulmonary hydatid disease may be pressure erosion of a bronchus or bronchial infection resulting from the obstruction. Further erosion into a branch arterial supply then causes hemoptysis. Hydatid cysts may erode into the great vessels, for example, the aorta, causing massive hemoptysis [11]. More common causes of pediatric hemoptysis include infection, foreign body aspiration, tracheostomy-related problems, congenital heart disease, cystic fibrosis, pulmonary hemosiderosis, trauma, and fictitious hemoptysis [12].

Radiological investigations are the mainstay for diagnosis in hydatid disease. CT scan differentiates between the fluid density of an intact cyst and ruptured cyst. Ruptured cysts show varied radiological appearances due to different combinations of the collapsed membrane, air, and fluid. However, the infected hydatid cyst may simulate a solid lesion due to an increase in attenuation levels. When positive history, serological tests, and other radiological signs are not present, complicated cysts seem to resemble 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 [7,13,14].

Laboratory testing should be used either in highly suspicious cases or for post-operative follow-up of pulmonary hydatid cyst disease. Serological tests are often helpful, but measurable immunological responses do not develop in some patients. The most sensitive technique in detecting pulmonary hydatid disease is immunoglobulin G enzyme-linked immunosorbent assay, with a sensitivity of 85.3%. Antibody production is elevated during the first 4–6 weeks after surgical intervention, followed by a decrease during the next 12–18 months. In patients have a recurrence before 2 years, antibody production remains similar to pre-operative levels [15,16].

The main treatment of a ruptured pulmonary hydatid cyst is surgery. Operative morbidity is higher in complicated pulmonary hydatid cysts than intact ones. The most frequent post-operative sequelae of complicated pulmonary hydatid cysts are prolonged air leakage (15.2%), empyema (9.1%), and atelectasis (9.1%). Medical treatment of pulmonary hydatid cysts with Albendazole or Mebendazole is used together with surgical treatment to avoid recurrence. It is given preoperatively 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 [17]. The usual dosage of orally administered Albendazole is 10–15 mg/kg/d in 2 divided doses, or a fixed dose of 400 mg twice a day. For Mebendazole, the daily dosage is 40–50 mg/kg in three divided doses [18]. The goal of surgical therapy in pulmonary hydatid disease is to remove the cyst while preserving as much lung tissue as possible. The surgical method may be different in intact (simple) and ruptured (complicated) cysts. Needle aspiration or enucleation can be attempted in cases of a simple cyst.

CONCLUSION

A possibility of hydatid cyst in cases of lung infections should be kept in endemic areas irrespective of the history and radiological features. Complete evaluation, along with tissue diagnosis, is important for establishing the confirmative diagnosis of tuberculosis. Empirical treatment with AKT, in cases of suspicion, should be avoided. The diagnosis should be revised at various steps in cases of suboptimal response hydatid cyst in the lung can get complicated and ruptured, if left untreated, and then cause mortality or morbidity. Serological tests may be of some help in cases of suspicion. A high index of suspicion for hydatid disease should be kept in children suffering from chronic lung diseases in endemic areas.

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