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Endobronchial Findings of Hydatid Cyst Disease: A Report of Five Pediatric Cases

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Summary. Hydatid disease is still an important public health problem throughout the world. Diagnosis of the disease is generally based on clinical and radiological findings. Evaluation of pulmonary disorders by flexible bronchoscopy (FOB) is a rapidly developing facility, but diagnostic and therapeutic FOB for pulmonary hydatid cysts is still controversial. This study examines the findings of endobronchial hydatid cyst disease in five pediatric patients from Turkey, and clinical experience about this subject is reviewed. All our patients presented with unusual symptoms of the disease, and for all of them, diagnosis had been delayed using current diagnostic methods. As a result of our experience, it can be reported that the endobronchial appearance of the hydatid cyst membrane is whitish-yellow, and it is difficult to differentiate it radiologically from some other common causes of endobronchial lesions in childhood, such as endobronchial tuberculosis, foreign body aspirations, mucous plaques, and granulation scars. The findings of these cases show that, hydatid cyst should also be kept in mind in differential diagnosis of endobronchial lesions. In the diagnosis of pulmonary hydatid cyst in children without typical clinical and radiological findings of the disease, FOB examination is a valuable diagnostic procedure. **Pediatr Pulmonol. 2012; 47:706–709.** © 2011 Wiley Periodicals, Inc.

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INTRODUCTION

Hydatid disease is a parasitic infestation caused by the larval form of the tapeworm *Echinococcus* (*E. granulosus*, and less commonly *E. multilocularis*). It is a major health problem in many parts of the world, with the incidence of infestation reported to be 2 per 100,000 in Turkey.[1–3] Even though infections may be acquired in childhood, because of the slowly growing nature of the echinococcal cyst, most cases of lung cysts become symptomatic and are diagnosed in adult patients. Only 10–20% of cases are diagnosed in patients younger than 16 years.[4] Most of the intact lung cysts are discovered incidentally on chest radiographs.[5] However, hydatid cysts may also rupture into the bronchial tree. Visualization of the membrane by flexible bronchoscopy (FOB) is uncommon in cases with incomplete expectoration. Evaluation of pulmonary disorders by FOB is a rapidly developing facility, but diagnostic and therapeutic FOB for pulmonary hydatid cysts is still controversial. Due to the risk of cyst rupture, most authors do not recommend bronchoscopy, except for excluding malignancy in adulthood, and for differential diagnosis of lung abscess, cavitary tuberculosis, bronchogenic cyst, and persistent pneumonic infiltrations in childhood.[6] There is little information in medical literature about the diagnosis of hydatid disease, using FOB evaluation, especially in childhood.[1] This article presents five pediatric cases from Turkey for each of whom a definitive diagnosis was made using FOB.

CLINICAL SUMMARIES

Patient 1

A 13-year-old boy was admitted to our clinic, with a 6-month history of cough and hemoptysis.[2] He had

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persistent infiltration in the right upper lung, which had proved resistant to 6 weeks of broad spectrum antibiotics therapy. The patient's history was unremarkable for tuberculosis in family members, contact with animals, or foreign body aspiration. On physical examination, breath sounds were significantly diminished over the upper right lung. Chest radiography showed a consolidation in the right upper zone and computerized tomography revealed a consolidation in the right upper lobe. Tuberculin skin testing was measured as 6 mm. FOB was performed for a differential diagnosis of non-resolving pneumonia and hemoptysis. The bronchoscopy showed a whitish endobronchial lesion totally occluding the orifice of the right upper bronchus (Fig. 1a). Since it would have been extremely difficult to remove with FOB, rigid bronchoscopy was performed. The lesion was extracted and pathological examination determined the typical three-layered structure of a hydatid cyst with an inner germinal layer, a middle acellular layer, and an outer fibrous capsule with eosinophils. An indirect hemagglutination test was positive for *Echinococcus granulosus*. No liver involvement was observed from ultrasonography. The patient was treated with albendazole, and complete clinical and radiological improvement was achieved in 2 months, with the treatment continuing for 6 months.

Patient 2

A 9-year-old girl, who had been taking albendazole for lung and liver hydatid cysts for more than 1 year, was admitted to our clinic with symptoms of chronic cough, sputum, recurrent pneumonia, and hemoptysis. Although the symptoms had been present for more than 6 months, an outside pediatric surgery clinic had stated that these complaints were not due to hydatid cyst disease, and she was referred to us to determine the underlying disease. Breath sounds were diminished over the lower left lung on physical examination. High resolution computerized tomography showed a 7 cm $\times$ 7 cm sized cystic lesion in the left lower lobe with adjacent atelectasis. Also, the cystic lesions on the liver and right lower lobe of lung were found to have reduced in size in comparison with previous tomography. To evaluate the atelectatic area on the left lower lobe, FOB was performed and the orifices of all the segments of the left lower lobe were found to be occluded with membranes of hydatid cysts (Fig. 1b). With the help of this procedure, a definite diagnosis of the disease could be made, and following excision of the cyst, the patient was totally asymptomatic.

Patient 3

A 5-year-old boy, who had a history of hemoptysis 3 weeks previously, had been treated with intravenous antibiotic for 10 days at another hospital. The infiltrations over the left lingulary lobe and the left upper lobe apical segment did not improve in spite of this treatment, and after the recurrence of hemoptysis in the second week of therapy, the patient was referred to our clinic for further evaluation. Other than the infiltrations mentioned above, lymphadenopathies, which were abnormally increased in size, were detected on computerized tomography. The patient's history was unremarkable for tuberculosis in family members and foreign

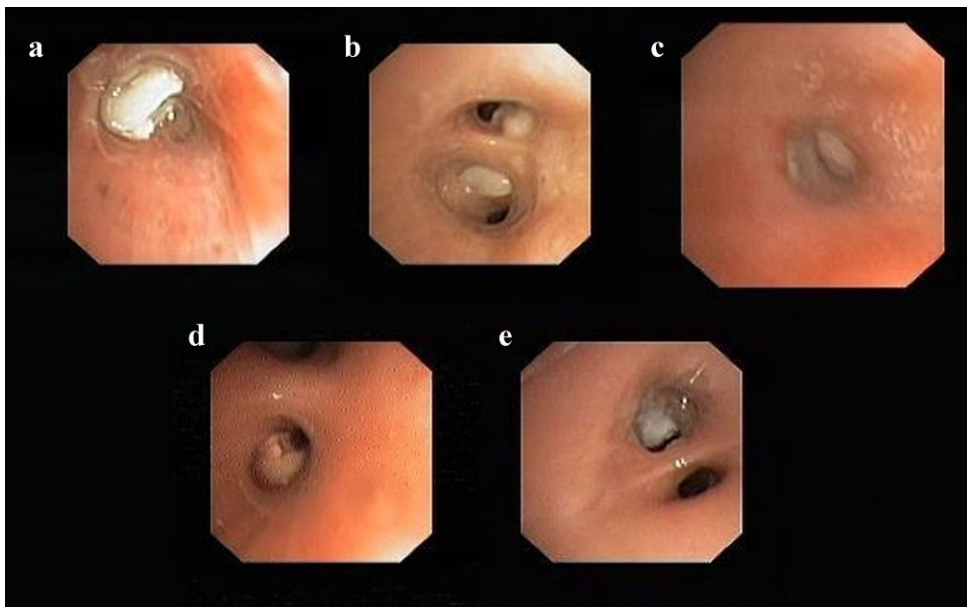

Fig. 1. Endoscopic appearance of the pulmonary hydatic cysts of the patients.

body aspiration. Bronchoscopy showed grayish endobronchial lesions totally occluding the orifices of the left lingulary lobe upper segment and left upper lobe apical segment (Fig. 1c). An indirect hemagglutination test was negative for *Echinococcus granulosus*. Tuberculin skin testing was measured as 2 mm, and ARB was determined as negative. However, due to the persistent infiltrations on chest X-ray after 4 weeks of antibiotic therapy, existence of lymphadenopathies on tomography, and endobronchial lesions which resembled tuberculosis, and keeping in mind that the patient lives in a country where tuberculosis is an endemic disease, anti-tuberculosis treatment was started with the consent of the parents. After 2 months of therapy, even though the patient was totally asymptomatic, the lesions persisted both on the X-ray and FOB. As hydatid cyst was suspected, we asked again about animal contact and it emerged that there was a history of contact with dogs in his neighborhood. The patient was retested for *Echinococcus granulosus* using an indirect hemagglutination test, and was determined as positive this time. Albendazole therapy was started, and complete clinical and radiological improvement was achieved in 2 months.

Patient 4

A 9-year-old girl presented with a 2-year history of hydatid cyst, which had been treated with albendazole for 6 months and followed-up at another hospital after the end of therapy. The patient had been experiencing symptoms of hemoptysis for 1 month, and albendazole treatment had been restarted 2 weeks before admittance to our clinic. Thoracic computerized tomography showed an image consistent with ruptured hydatic cyst on the left lower lobe. Due to the persistent hemoptysis, we performed a bronchoscopy, and during the procedure, fresh bleeding from the orifice of the left lower lobe was detected. After suctioning of the blood, whitish hydatid cyst membrane was seen which was totally occluding the orifice of the lobe (Fig. 1d). The patient underwent lobectomy because of massive hemoptysis, and albendazole therapy was continued for a further 6 months.

Patient 5

A 13-year-old boy was admitted to the clinic with a history of hydatid cysts on the right lower and left upper lobes 2 years previously, which had been treated with albendazole for 3 months. The first diagnosis was said to be not definite by an external clinic so the therapy was halted after 3 months, and there had been no follow-up since then. The patient had been asymptomatic from that time until 2 weeks prior to admittance when symptoms of respiratory distress and fever had begun and continued over 2 weeks. Chest examination revealed a decrease in lung sounds over the right lower area and radiographs showed findings consistent with a ruptured hydatid cyst 8 cm $\times$ 8 cm in size, with air-fluid level in the right lower region and pleural effusion in the adjacent area. The differential diagnoses made by the radiologists were lung abscess, ruptured hydatid cyst, and bronchogenic cyst. A serum sample for IHA was obtained, and to evaluate the endobronchial structures, bronchoscopy was performed. FOB revealed a membranous image, which was occluding the orifice of the posteriobasal segment of the right lower lobe (Fig. 1e). An indirect hemagglutination test was positive for *Echinococcus granulosus*. Albendazole treatment was restarted, and after 6 months of therapy, complete clinical and radiological improvement was achieved.

DISCUSSION

This study examines the findings of endobronchial hydatid cyst disease in five pediatric patients, and clinical experience about this subject is reviewed. Hydatid disease is still an important public health problem throughout the world. Diagnosis of the disease is generally based on clinical and radiological findings. Clinical symptoms usually develop because of one or more of these three factors: (a) Mechanical pressure or deformation of the tissues or nearby vascular structure, (b) rupture or leakage of the cyst, or (c) superinfection.[5,6] Cough, hemoptysis, and chest pain are non-specific findings for the disease.[7,8] Intact pulmonary cysts are frequently detected by chest radiography, and typically appear as solitary or multiple well-defined, round, opaque lesions. However, the radiological appearance may become atypical and it may cause incorrect and delayed diagnosis when a hydatid cyst is infected or ruptured.[9,10] These kinds of complications can imitate several pleural and pulmonary diseases such as non-resolving pneumonia, tuberculosis, abscess, and tumor.[10–12]

FOB is unnecessary in patients with a typical clinical and radiological picture, but it can be performed for a differential diagnosis in cases of atypical radiological appearance, when there is a suspicion of tumor, and to trace the source of hemoptysis.[1,11,13] FOB was performed on the cases in this study because of symptoms of hemoptysis and cough in four patients, and because of persistent respiratory distress in one patient. Atypical and persistent radiologic abnormalities had also been observed in all the patients. With the use of FOB, a section of membrane can be taken for biopsy analysis and examined to confirm its laminated nature, or the bronchial aspirate can be analyzed for the presence of hooklets.[13] Specific and non-specific bronchoscopic findings for pulmonary hydatid cysts have been

described in adults.[1,2,11] The appearance of whitish-yellow laminated membrane in a bronchus draining the cyst cavity is diagnostic for the disease.[13–15] However, there is little information in literature about the bronchoscopic findings of hydatid disease in childhood.[2,12,15] As a result of our experience, it can be reported that the endobronchial appearance of the hydatid cyst membrane is whitish-yellow, and it is difficult to differentiate it radiologically from some other common causes of endobronchial lesions in childhood, such as endobronchial tuberculosis, foreign body aspirations, mucous plaques, and granulation scars. Especially in developing countries, tuberculosis is still an endemic disease, and the appearance of an endobronchial lesion during FOB may sometimes mislead to a diagnosis of tuberculosis, which may result in using lengthy anti-tuberculosis therapy and delay the actual disease diagnosis. The findings of these cases show that, hydatid cyst should also be kept in mind in differential diagnosis of endobronchial lesions. Medical therapy of hydatid disease is the best therapy except in cases of large pulmonary cysts and when complications occur, such as compression of parenchyma, obstruction of airways, or massive hemoptysis.[7,8] Of the cases reported in this article, three were successfully treated with oral albendazole, one underwent lobectomy due to uncontrollable hemorrhage bleeding, and one underwent surgery as the large cyst was causing airway compression symptoms.

To the best of our knowledge, this is the largest hydatid cyst disease pediatric case series in which FOB was used for diagnosis and follow-up. All our patients presented with unusual symptoms of the disease, and for all of them, diagnosis had been delayed using current diagnostic methods. In the diagnosis of pulmonary hydatid cyst in children without typical clinical and radiological findings of the disease, FOB examination is a valuable diagnostic procedure. When endobronchial whitish lesions are detected during bronchoscopies for differential diagnosis of lung diseases or for any other reason, hydatid cyst disease should also be considered as the possible diagnosis. A larger case series may better define the role of FOB for diagnosis and follow-up of pulmonary hydatic cysts in children.

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