Fibreoptic bronchoscopy and bronchoalveolar lavage for confirmation of pulmonary hydatid cyst.

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June 22, 2020

Abstract

There is no confirmatory diagnostic test for pulmonary hydatid cyst other than surgical excision and histopathologic confirmation. Imaging is at best suggestive and serology does not have a satisfactory sensitivity. We present a series of children wherein flexible fibreoptic bronchoscopy under conscious-sedation, revealed hydatid membranes in airways. Broncho-alveolar lavage analysis confirmed hydatid in half of them. We propose flexible fibreoptic bronchoscopy with broncho-alveolar lavage as a confirmatory diagnostic test for pulmonary hydatid in children. To the best of our knowledge, this is a completely novel approach to the condition with potential to alter the diagnostic paradigm.

Introduction

Hydatid disease is a worldwide zoonosis, but endemic in some regions. Cystic hydatid disease or hydatidosis is caused by the cestode*Echinococcus granulosus* , whereas alveolar hydatid disease is caused by *Echinococcus multilocularis* . The cestode life cycle requires a definitive host that harbours the mature parasite and an intermediate host for the immature stage of the parasite. Humans are accidental dead end hosts, who acquire the eggs via the feco-oral route.

The liver is most often affected in adults, whereas lungs are more frequently involved in children[1]. The clinical presentation depends on cyst site, size and complications like infection. In more than one-third cases, the condition can be asymptomatic[2]. Symptomatic children develop cough, chest pain, shortness of breath, expectoration, fever and hemoptysis. The diagnosis can be suspected on the basis of endemicity, history of exposure to sheep or dogs, chest imaging showing a single or multiple cysts and positive serology test. Uncomplicated pulmonary cysts appear as homogenous radio opacities on chest xray and well circumscribed fluid attenuation lesions on CT. Complicated cysts sometimes have appearances described as crescent sign, cumbo sign, water lily sign, etc., however, there may be atypical radiologic signs. Chest ultrasonography may detect peripheral lesions. ELISA for IgG antibodies has sensitivity of 85-98% for liver cysts, but only 50-60% for lung cysts, thereby creating false positive and false negative reports[3]. Percutaneous aspiration of the cyst carries the risk of rupture and an anaphylactic reaction. Thus, there is no established method to confirm the diagnosis and this can be done only by pathologic examination of surgically excised cysts.

Here we report a series of eight children with suspected and unsuspected lung hydatid who underwent bronchoscopy and bronchoalveolar lavage (BAL) under conscious sedation. We were able to confirm hydatid disease in seven children using this novel approach.

Methods

The cohort comprised eight children investigated for persistent respiratory symptoms and/or persistent radiological abnormalities. Hydatid cyst was considered as a differential diagnosis in five of them.

A complete blood count and differential was done in all children. Chest xrays and CT films were reviewed by a Pediatric Radiologist. Abdominal ultrasonography was used to screen for extrapulmonary cysts. An IgG titre of 1:800 on ELISA was considered positive hydatid serology.

All children underwent flexible fibreoptic bronchoscopy (FFOB) under conscious sedation using a 3.6 mm or 4.8 mm scope. Prior written, informed consent was obtained from a parent, as per the institutional protocol. Sedation was administered using oral triclofos (50 mg/kg) and intravenous midazolam (0.1 mg/kg). Bronchoscopy findings were recorded and BAL fluid was examined. The sediment of centrifuged fluid was also examined. Whenever possible, aspirated material was also sent for histopathologic examination. No lung biopsies were taken during the procedure.

Results

Among the eight children, five were boys. The age ranged from 6 to 12 years. Cough was present in seven children; it was dry in four and wet or productive in three cases. Fever was present in five children. Other symptoms were chest pain, hemoptysis and dyspnea. On examination, localized chest findings were present in six children. Five of them had reduced breath sounds with dullness on percussion, while one had bronchial breathing. One child had been treated as empyema 2 years back in another hospital, and another child had received treatment for lung abscess. They presented to our hospital because of non-resolution. Table 1 presents a summary of the clinical findings.

All children presented to us with chest xrays and CT scans done elsewhere. Xrays showed cystic lesion in one patient, homogenous consolidation with rounded contours in two and non-specific localized consolidation in the other five cases. CT scan showed fluid filled cystic lesions in two, while the others showed consolidation with or without central cavitation. One child also had a right paratracheal cystic lesion. The radiographic findings are summarized in Table 1. Four children had an eosinophil count above 500 per microlitre. Hydatid serology was positive in six children. One child had an additional cyst in segment V of the liver.

Flexible fibreoptic bronchoscopy showed white or yellow glistening membranes occluding the main, lobar or segmental bronchi, in all but one patient. The location corresponded to the involved lobe identified on imaging. BAL specimens were carefully obtained in seven children.

There were no systemic adverse events during any of these procedures, including hemodynamic instability, anaphylactic reaction or other post procedure complications. However, in one child, there was a gush of fluid return during BAL that turned mildly haemorrhagic and required prolonged suctioning. No other intervention was required in this child.

Cytopathologic examination of seven BAL specimens showed acellular lamellated membranes in two specimens, and non-specific inflammatory cells in the remaining five. Membranes were brightly positive on Periodic acid-Schiff (PAS) stain. Two BAL specimens had solid fragments wherein histopathologic examination showed acellular eosinophilic lamellated membrane representing hydatid ectocyst in both. Hydatid germinal layer could also be seen in one of them. Cytologic examination in these two specimens did not show any hydatid elements. Thus pathologic confirmation was obtained in four of seven BAL specimens.

Seven children were referred for surgery and histopathology of the excised cysts confirmed hydatid disease. The patient who did not have membranes on bronchoscopy had a normal BAL examination. This child had positive serology and was treated for hydatid without surgery.

Bronchoscopy showed membranes in two children with negative serology. Overall, flexible bronchoscopy with BAL analysis could confirm the diagnosis in seven of eight children patients prior to surgery.

Discussion

Currently, a clinical diagnosis of pulmonary hydatid is based on chronic respiratory symptoms, supported by compatible radiologic findings and hydatid serology. However, imaging findings in a complicated cyst can overlap with lung abscess, tuberculosis, tumor, Wegener's granulomatosis, bronchiectasis, pneumothorax or empyema. Serology in pulmonary haydatidosis has lower sensitivity than hepatic hydatidosis. It can be false positive in other helminthic infections, cancer, chronic immune disorders, liver cirrhosis, presence of anti P1 antibodies, etc; and may be false negative if the cyst is unruptured[3]. Thus it would be helpful if better diagnostic modalities are available.

There is no consensus regarding the place of bronchoscopy in diagnosis. To the best of our knowledge, this is the first series reporting FFOB under conscious sedation, in pediatric lung hydatid.

However, multiple reports in adult patients documented the unexpected visualization of hydatid membranes during procedures performed for persistent respiratory symptoms or radiological shadows. In a retrospective analysis of 386 cases of lung hydatid, bronchoscopy performed prior to surgery in 106 patients helped in establishing the diagnosis in 21 patients[4]. In another retrospective study of 72 patients with pulmonary hydatid, bronchoscopy performed in 34 patients showed cyst membranes in 7 patients[5]. Other non-specific findings described by the authors included airway hyperemia, edema, purulent secretions and extrinsic compression. The relatively higher diagnostic yield in our series could be because most children had complicated cysts.

Our experience suggests that FFOB with BAL (performed under conscious sedation) could be an effective diagnostic modality, especially since radiology findings or serology may be inconclusive even in suspected cases. However, the procedure should be performed with great caution, with preparedness for managing complications.

Authors' contribution:

KK participated in clinical care of patients, and pre as well as post bronchoscopy monitoring. He drafted the manuscript.

JLM participated in clinical care of patients, performed bronchoscopy and BAL in some of the children, corrected and finalized the manuscript. He will act as guarantor.

PV participated in clinical care of patients, performed bronchoscopy and BAL in some of the children.

AB performed histopathologic examination on two BAL specimens containing solid fragments, and all surgically excised cysts.

NG performed cytopathologic examination of seven BAL specimens.

KKS participated in clinical care of patients, and pre as well as post bronchoscopy monitoring.

AS examined and reviewed the radiologic images of the children.

MS is the Head of the Pediatric Pulmonology Unit, supervising all clinical care of the children in the Unit.

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