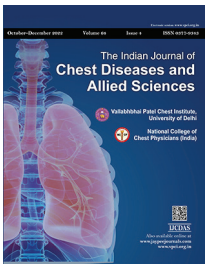
CASE REPORT

"Air Bubble Sign" and Fibreoptic Bronchoscopy in Complicated Pulmonary Hydatid Cysts: Steps in the Right Direction



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Abstract

Intact pulmonary hydatid cysts are often discovered incidentally on chest X-rays (CXRs) as rounded opaque lesions with smooth borders. Cyst rupture and infection (complicated hydatid cyst) often change the radiologic appearance delaying diagnosis. Air bubble sign on computed tomography is recognized as a sign of high sensitivity and specificity in the diagnosis of complicated hydatid cysts when compared to other classical radiologic signs. Bronchoscopy is not a preferred investigation in patients with intact hydatid cysts but may have a major role in patients with complicated hydatid cysts with atypical presentation. In the present case series, we attempt to highlight the importance of the "air bubble sign" and the identification of a whitish membrane at bronchoscopy in the diagnosis of complicated hydatid cysts.

Keywords: Air bubble sign, Bronchoscopy, Pulmonary hydatid cyst.

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Abbreviations used in this Article

CT = Computed tomographic; CXR = Chest X-ray; FOB = Fibreoptic bronchoscopy.

Key Message

"Air bubble sign" on the computerized tomography and visualization with bronchoscopy of a whitish membrane in the involved bronchus can often clinch a diagnosis in an otherwise complicated case of hydatid cysts with an atypical presentation.

Introduction

Pulmonary hydatid cysts are not infrequent in clinical practice in a developing country like India. The cysts are often discovered incidentally on CXR as homogeneous oval masses with smooth borders surrounded by normal lungs.[1] Cyst rupture with subsequent infection (complicated hydatid cyst) often prompts symptoms and medical attention. Though diagnostic radiologic signs of cyst rapture like the "crescent sign" or "water lily" sign have been described, they are not that common.[1] More frequently, complicated hydatid cysts resemble a lung mass, abscess or nonresolving pneumonia on imaging, and delayed diagnosis.

Though bronchoscopy is not warranted in intact hydatid cysts, it helps in the workup of complicated hydatid cysts with an atypical presentation. Here, we present four cases highlighting the potential importance of fibreoptic bronchoscopy (FOB) and a not so well-known "air bubble sign" in the diagnosis of complicated hydatid cysts.[2,3]

Case Description

Case 1

A 55-year-old housewife, a nonsmoker, presented with a 9-month history of cough and frequent blood streaking of the sputum. There was no clubbing or lymphadenopathy. CXR showed opacity in the right mid zone and a thoracic computed tomographic (CT) scan showed a large (43 × 62 mm) well-defined, heterogenous density mass lesion in the apical segment of the right lower lobe. CT-guided trucut biopsy of the lesion (Fig. 1), done on two occasions, was noncontributory. On FOB, the right apical segment bronchus opening was edematous with purulent secretions. Bronchial lavage was negative for malignant cells. A long history in an otherwise-well patient with inconclusive investigations convinced the surgeon to go for an exploratory thoracotomy. During the operation, the diagnosis became clear with histopathology confirming the final diagnosis as a complicated hydatid cyst.

Case 2

A 45-year-old lady, nonsmoker, complained of a cough with scanty sputum for 10 weeks. There were occasional hemoptysis and right-sided chest pain for the last 6 weeks. There were a dull note and diminished breath sound in the right mammary region. CXR showed the right lower zone opacity, and contrast-enhanced CT scan of the thorax reported "a solid, well-defined, heterogeneously enhancing mass lesion in the right middle lobe." Ultrasound-guided core biopsy of the lesion was suggestive of interstitial fibrosis and

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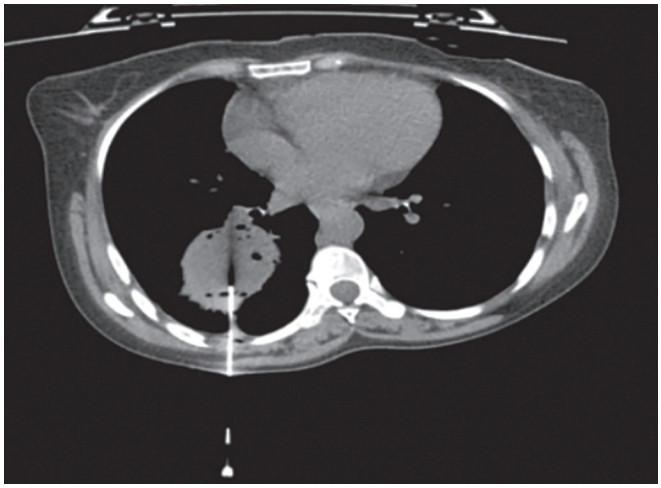


Fig. 1: CT guided biopsy being taken from the heterogenous density mass lesion showing "air bubble sign", case 1

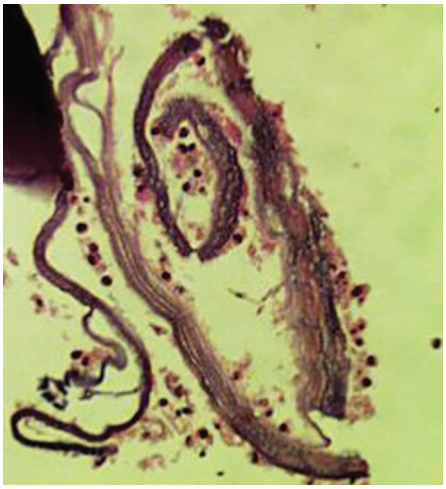


Fig. 2: Laminated membrane suggestive of hydatid cyst as seen on Hematoxylin and Eosin staining, case 2

inflammation. On bronchoscopy, there was a whitish material/tissue overlying the right middle lobe bronchus orifice. We were surprised when the biopsy revealed a laminated membrane suggestive of a hydatid cyst (Fig. 2).

Case 3

A 33-year-old woman, non-smoker, normoglycemic, presented with a one-year history of cough, scanty expectoration, and hemoptysis. There was no clubbing or lymphadenopathy and sputum was negative for acid-fast bacilli. High-resolution computed tomography (HRCT) scan suggested lateral basal segmental consolidation of the left lower lobe. On FOB, a whitish membrane-like material protruding from the lateral basal segment of the left lower lobe bronchus was observable (Fig. 3). Bronchial lavage showed fragments of a lamellar membrane and scolices and the biopsy also revealed the lamellar membrane of hydatid disease.

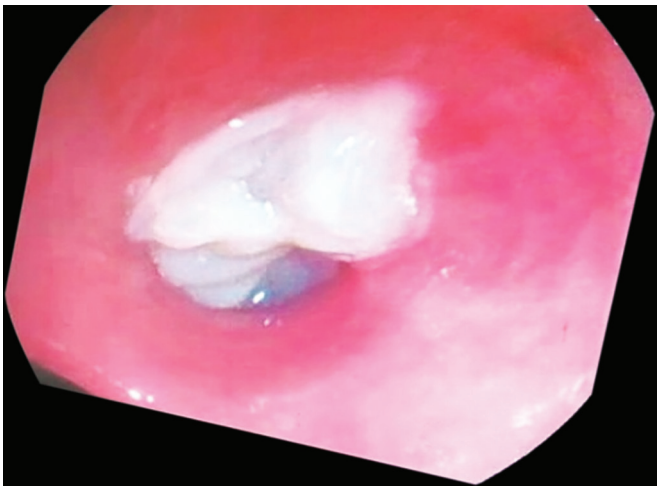
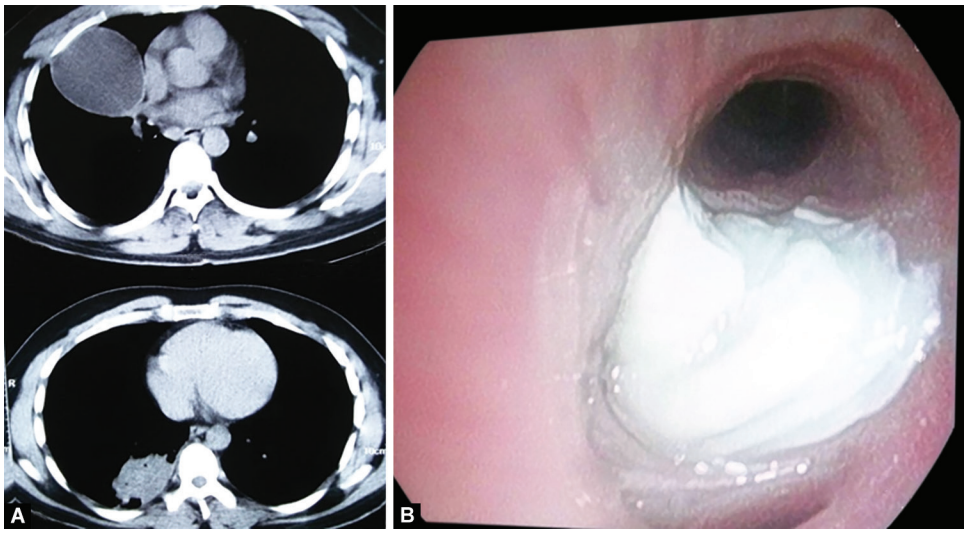


Fig. 3: Whitish membrane-like material protruding from the lateral basal segment of the left lower lobe bronchus, case 3



Figs 4A and B: (A) CT thorax showing a cystic lesion in the right middle lobe and an irregular heterogeneous mass-like lesion in the posterior basal segment of the right lower lobe showing "air bubbles," Case 4. (B) Whitish crumpled tissue-like material was found at the orifice of the posterior basal segment of the right lower lobe, case 4

Case 4

A 35-year-old farmer, smoker, presented with a persistent cough for a year, a history of expectoration 9 months back of a clear salty-tasting fluid for a day, and also an episode of hemoptysis. CT thorax showed a cystic lesion in the right middle lobe and an irregular heterogeneous mass lesion in the posterior basal segment of the right lower lobe having a couple of "air bubbles" (Fig. 4A). With a classical history and a typical intact cyst in the middle lobe, we went for bronchoscopy to evaluate the second lesion. A whitish crumpled tissue-like material was found at the orifice of the posterior basal segment (Fig. 4B). Bronchial lavage was noncontributory, but the biopsy showed a lamellated membrane of hydatid cyst.

Our four patients underwent thoracotomy for cyst removal with uneventful recovery.

Discussion

Hydatid disease is caused by the ingestion of eggs acquired from contact with dogs infested with the adult worm of *Echinococcus granulosus*. The eggs develop into larvae in the duodenum, penetrate its wall, and pass into the portal bloodstream to lodge in the capillary beds of the liver or lungs where only a few survive and develop into cysts. Approximately 70% of the cysts occur in the liver and 10–30% in the lungs.[1] Multiple lung cysts (in 30%) or coexisting liver cysts (20–40%) help in the diagnosis.

The cyst itself has two layers: an outer protective acellular laminated layer, the exocyst, and an inner germinal layer, the endocyst. The germinal layer produces the intracystic fluid and gives rise to the brood capsules/daughter cysts within which larval scolices develop. The cyst itself is surrounded by a pericyst, a host-derived adventitial layer of fibrous tissue. Cyst rupture and infection often give rise to symptoms: cough, hemoptysis, chest pain, and fever. Expectoration of salty fluid from cyst rupture into the airways is rarely elicited (Case 4). Following the cyst rupture, the detached membranes may float on the residual fluid in the pericyst giving rise to classic radiologic signs like "water lily" or "camelot."[1] With a breach of the pericyst only, air may enter between the pericyst and the endocyst producing the "meniscus" or "air crescent" sign. Though diagnostic, these findings are uncommon.

CT demonstrates water-density lung cysts and can also identify detached or collapsed membranes and daughter cysts helping in diagnosis. However, complicated hydatid cysts often have higher CT attenuations[4] due to hemorrhage, infection, or fibrosis, so they can be confused with solid neoplasms, abscess, or cavitary lesions as found in our cases, prompting trucut biopsy in two cases (Cases 1 and 2). The presence of "air bubbles" seen as single or multiple small, rounded, radiolucent areas with sharp margins in the periphery of a solid mass lesion, best seen in the mediastinal window, has high sensitivity and specificity (83.1% and 94.5%, respectively) in establishing the diagnosis of a complicated hydatid cyst.[2] This sign is thought to be due to the dissection of air between the pericyst and ectocyst. This sign is present in the right lower lobe lesion in Case 4 and was missed during the initial workup of Case 1.

Suspicion of malignancy indicated bronchoscopy in the first three of our cases. In Case 2, at bronchoscopy, the whitish friable material biopsied turned out to be a hydatid cyst membrane. In Cases 3 and 4, with the visualization of the whitish membrane fragment at bronchoscopy, we knew that we were dealing with a ruptured hydatid cyst. Looking up literature, we found several case reports of hydatid cysts being diagnosed in a similar manner.[5,6] There have been a few studies from Turkey where FOB was able to make a diagnosis in more than 50% of cases of complicated pulmonary hydatid cysts.[7,8]

Surgical removal being the main therapeutic option, a preoperative diagnosis helps in planning the surgery and is a relief to the patient and the surgeon in knowing that they are not dealing with a neoplastic lesion.

Therefore, to conclude, the diagnosis of complicated pulmonary hydatid is sometimes challenging. Along with various radiologic signs, recognition of the "air bubble sign" is often a step in the right direction. Visualization of the hydatid membrane at bronchoscopy is a rewarding experience, giving us an instant working diagnosis.

References

1. Morar R, Feldman C. Pulmonary echinococcosis. Eur Respir J 2003; 21(6):1069–1077. DOI: 10.1183/09031936.03.00108403.
2. Köktürk O, Oztürk C, Diren B, et al. "Air bubble": a new diagnostic CT sign of perforated pulmonary hydatid cyst. EurRadiol 1999;9: 1321–1323. DOI: 10.1007/s003300050840.
3. Yuncu G, Ors Kaya S, Sevinc S, et al. The diagnostic value of the 'Air Bubble Sign' in complicated pulmonary hydatid cysts. J Thorac Cardiovasc Surg 2007;133:1524–1574. DOI: 10.5580/1676.
4. Koul PA, Koul AN, Wahid A, et al. CT in pulmonary hydatid disease: Unusual appearances. Chest 2000;118(6):1645–1647. DOI: 10.1378/chest.118.6.1645.
5. Basu A, Dhamija A, Agarwal A, et al. Ruptured pulmonary hydatid disease mimicking a lung mass: Diagnosed by flexible video bronchoscopy. BMJ Case Rep 2012;2012:bcr2012006977. DOI: 10.1136/bcr-2012-006977.
6. Madan K, Singh N. Bronchoscopic diagnosis of pulmonary hydatid cyst. CMAJ 2012;184(2):E158. PMID: 22105751.
7. Saygi A, Oztek I, Guder M, et al. Value of fiberoptic bronchoscopy in the diagnosis of complicated pulmonary unilocular cystic hydatidosis. Eur Respir J 1997;10(4):811–814. PMID: 9150317.
8. Ulaş AB, Eroğlu A, Aydın Y, et al. The diagnostic value of fiberoptic bronchoscopy in ruptured lung hydatid cysts.Turk Gogus Kalp Damar Cerrahisi Derg 2019;27:350–354. DOI: 10.5606/tgkdc.dergisi.2019.17479.