Unusual presentation of more common disease/injury

Ruptured pulmonary hydatid disease mimicking a lung mass: diagnosed by flexible video bronchoscopy

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Summary

Echinococcosis is frequently seen in an endemic country like India. The vast majority of infestations in humans are caused by *Echinococcus granulosus*. The cyst is most commonly seen in the liver in about 74% of the cases, followed by pulmonary involvement in about 24% of cases. In our case report, a 25-year-old man presented with a mass-like lesion in the upper and middle lobe of the right lung. The patient had deteriorated clinically as the mass like opacity grew in size. Here, we have highlighted an uncommon presentation of hydatid disease of the lung diagnosed by video bronchoscopy and bronchoalveolar lavage (BAL) as clinico-radiological examination along with FNAC (fine-needle aspiration cytology) of the lesion proved insufficient tools to make a diagnosis.

BACKGROUND

Echinococcosis or hydatid disease, caused by larvae of the tapeworm *Echinococcus*, is frequently seen in an endemic country like India. The vast majority of infestations in humans are caused by *Echinococcus granulosus* which causes unilocular cysts while *Echinococcus multilocularis* is rare and causes multilocular cysts. The cyst is most commonly seen in the liver in about 74% of the cases, followed by pulmonary involvement in about 24% of cases. The peritoneum, kidney, brain and mediastinum are involved individually in 3–4% of the cases, while the heart is involved in 1–2% of the cases. Other organs notably the bone, spinal cord, pleura, bladder, ovary and thyroid gland are involved individually in less than 1% of the cases.^{1,2} Isolated pulmonary involvement is uncommonly seen in hydatid disease. Pulmonary hydatid cyst is more common on the right side especially in the lower lobes but it can occur in any lobe of the lung.³ A complicated hydatid cyst is defined as a cyst which has ruptured into the bronchus or pleural cavity with or without infection.⁴

CASE PRESENTATION

A 25-year-old man, a labourer by occupation, non-smoker with no other addiction and no other comorbid illness presented to us with chief complaints of fever, cough with expectoration and right-sided chest pain for 10 months along with haemoptysis for 6 months. On examination, the patient was found to be pale with blood pressure of 100/60 mm Hg, pulse rate of 110/min, respiratory rate of 24/min and a body temperature of 100.6°F. On examination of the chest bronchial breath sounds were present in right infraclavicular area along with fine crepts. Blood examination revealed a Hb%—9 g/dl, total lymphocyte count (TLC)—12 000/mm³, platelet—3.68 lakh/mm³ with a normal liver function test and a normal renal function test. Chest x-ray of the patient

revealed an inhomogenous opacity in the right upper and middle lung zones (figure 1). The lesion was initially small, located in the right paratracheal area which had progressively increased over a period of 10 months. The patient had deteriorated clinically as the mass like opacity grew in size. In the last 10 months, the patient had received amoxycillin-clavulanate for about 2 weeks followed by cefoperazone-sulbactam and levofloxacin for about 3 weeks. This was followed by an 8-month course of antitubercular treatment (3 months of rifampicin, isoniazid, ethambutol and pyrazinamide followed by 5 months of rifampicin and isoniazid) with no response. Ultrasound of the abdomen was normal. Mantoux test of the patient was also negative. Sputum examination did not reveal any acid-fast bacilli (AFB) or malignant cells. Contrast-enhanced CT (CECT) thorax of the patient revealed an inhomogenous mass-like lesion in the right upper and middle lobe with areas of necrosis. Enlarged mediastinal lymphadenopathy (including right lower paratracheal and right hilar lymph nodes) was present. There was no pleural effusion associated with the lesion. CT-guided fine-needle aspiration cytology (FNAC) of the lesion was carried out, which did not reveal any malignant pathology and the procedure was not associated with any complication. It seemed to be a stage IIIA lung mass with non-bulky mediastinal lymphadenopathy and hence positron emission tomography (PET) scan was carried out to assess the possibility of neoadjuvant therapy followed by surgical resection in the patient. PET scan of the lesion revealed fluoro deoxyglucose (FDG)-avid areas (SUV of 12.1) in the periphery of the lesion (figure 2) along with FDG-avid mediastinal lymphadenopathy (SUV of 5.3). Endobronchial ultrasound (EBUS) FNAC of the mediastinal lymphadenopathy was carried out which showed reactive lymph node enlargement with no evidence of malignancy (figure 3). The patient was then subjected to video bronchoscopy which revealed rounded whitish opacities in the

Figure 1 Preoperative chest x-ray of the patient showing a small right paratracheal opacity initially which increased to occupy the right upper and middle lobes of the lung.

apical and anterior segmental openings of the right upper lobe (figure 4). These whitish opacities had a laminated membrane covering them. Bronchial washings were taken from the involved segments in the right upper lobe. A portion of the white laminated membrane was also sucked out during aspiration of bronchial washings. Wet-mount examination of the bronchial washings revealed hooklets of hydatid. Broad branched septate hyphae were also seen in the bronchial washings which on culture revealed the growth of *Aspergillus fumigatus*. Galactomannan test in the serum for *Aspergillus* was also positive. The laminated membrane covering the opacities revealed germinal epithelium suggestive of hydatid. Serology for hydatid was performed in the serum after the demonstration of hooklets and the germinal epithelium which was also positive. The patient

was negative for HIV, hepatitis B and hepatitis C with no other evidence of immunosuppression.

The patient was finally diagnosed as a case of ruptured pulmonary hydatid cyst with *A fumigatus* superinfection. The patient was treated with albendazole and voriconazole for 11 days preoperatively. As the patient's general condition improved he was subjected to right upper and middle lobectomy. The patient was taken for an elective right bilobectomy (right upper and middle lobectomy) by the conventional fourth intercostal right posterolateral thoracotomy along with mediastinal lymphadenectomy (figure 5). Superior pulmonary vein was divided by Endo-GIA 45 mm stapler using a white load. The main fissure was developed and the branches of pulmonary arteries were also divided using white load. Ultimately,

Figure 2 Positron emission tomography CT thorax showing increased fluoro deoxyglucose uptake by the lesion.

Figure 3 Endobronchial ultrasound fine-needle aspiration cytology from the mediastinal lymphadenopathy.

both the upper and middle lobar bronchus were taken up and divided separately by Endo-GIA 60 mm using green load. The remaining lung parenchyma was divided by blue load (J&J). Haemostasis was achieved after thoroughly cleaning the cavity with hypertonic saline. Two 28-french intercostal drains were placed in the pleural cavity and the chest was closed in layers (figure 6). The resected specimen on histopathological examination also revealed germinal epithelium (figure 7). The intercostal drains were removed on the eighth postoperative day. Postoperatively, albendazole and voriconazole were continued and the patient has now been on treatment for the last 2 months with significant improvement.

INVESTIGATIONS

Blood examination revealed a Hb%—9 g/dl, TLC—12 000/mm³, platelet—3.68 lakh/mm³ with a normal liver function test and a normal renal function test. Chest x-ray of the patient revealed an inhomogenous opacity in the right upper and middle lung zones. The lesion was

initially small, located in the right paratracheal area which had progressively increased over a period of 10 months. The patient had deteriorated clinically as the mass-like opacity grew in size. Ultrasound of the abdomen was normal. CECT thorax of the patient revealed an inhomogenous mass-like lesion in the right upper and middle lobe with areas of necrosis. Enlarged mediastinal lymphadenopathy (including right lower paratracheal and right hilar lymph nodes) was present. There was no pleural effusion associated with the lesion. PET scan of the lesion revealed FDG-avid areas (SUV of 2.1) in the periphery of the lesion along with FDG-avid mediastinal lymphadenopathy. The lesion was reported as 'likely mitotic' on PET CT thorax. CT-guided FNAC of the lesion was carried out which did not reveal any malignant pathology. Mantoux test of the patient was also negative. Sputum examination did not reveal any AFB or malignant cells. The patient was then subjected to video bronchoscopy which revealed rounded whitish opacities in the apical and anterior segmental openings of the right upper lobe. These whitish opacities had a laminated membrane covering them. Bronchial washings were taken from the involved segments in the right upper lobe. A portion of the white laminated membrane was also sucked out during aspiration of bronchial washings. Wet-mount examination of the bronchial washings revealed hooklets of hydatid. Broad branched septate hyphae were also seen in the bronchial washings which on culture were suggestive of coexistent *A fumigatus* superinfection. The laminated membrane covering the opacities revealed germinal epithelium suggestive of hydatid.

Figure 4 Video bronchoscopy revealed rounded whitish opacities covered by laminated membrane in the anterior and apical segments of the right upper lobe.

Figure 5 Surgically resected specimen of right upper lobe and right middle lobe.

Figure 6 Postoperative chest x-ray after right upper and middle lobectomy and placement of muscle flap.

DIFFERENTIAL DIAGNOSIS

- ▶ Pulmonary tuberculosis
- ▶ Bronchogenic carcinoma
- ▶ Bacterial pneumonia
- ▶ Fungal pneumonia

TREATMENT

The patient was treated with albendazole and antifungals. Later, lobectomy of the right upper and middle lobes was performed in our case.

OUTCOME AND FOLLOW-UP

Two 28-French intercostal drains were placed in the pleural cavity and the chest was closed in layers. The intercostal drains were later removed on the eighth post-operative day and the patient was discharged in a stable condition.

Figure 7 Histopathological examination of the removed laminated membrane showing the presence of germinal epithelium.

DISCUSSION

Lewell⁵ has classified hydatid cysts into three types according to their imaging appearance. Type I is a fluid-filled cystic structure. Type II is the hydatid cyst with presence of daughter cysts and/or matrix. Type III is a mummified or inert calcified lesion. Hydatid disease of the lung is easier to diagnose in its typical presentation but in its atypical presentation it is often difficult to diagnose. Through this case we have presented a rare presentation of the lung hydatid as a lung mass occupying the right upper lobe and middle lobe. At a first glance it seemed to be a very slow growing tumour of the lung which had gradually increased over the past few months. CT-guided FNAC was negative for malignancy and PET CT thorax revealed FDG-avid areas in the periphery of the lesion. There is usually no increased FDG uptake by the intact hydatid cyst⁶ but in case of a ruptured hydatid cyst there may be increased FDG uptake especially in the periphery of the hydatid cyst.⁷⁻⁹ Video bronchoscopy is usually required in patients with atypical presentation of the hydatid disease¹⁰⁻¹² as in our case. Serology for hydatid was positive in the serum. The overall sensitivity of ELISA for serodiagnosis of hydatid is about 96%^{13 14} but these serodiagnostic techniques have been shown to give false results. Radiography including CT scan thorax is an accurate diagnostic test for typical presentation of the hydatid disease. Simple cysts are homogeneous with a fluid(water) density and a thin enhancing rim with contrast whereas infection of the cyst may increase the attenuation values.¹⁵ The ruptured membranes of hydatid cyst have been described as 'crescent', 'water-lily', 'double arch or cumbo's' sign, 'ring within ring', 'mass within a cavity', 'serpent' or 'snake' and 'spin' or 'whirl' sign.¹⁶ Infected hydatid cysts may present with air bubbles within the cyst.¹⁷ Unintentional fine-needle aspiration (FNA) of pulmonary hydatid cyst has been reported on several occasions in order to obtain material for diagnosis of suspected mass lesion without any complications such as anaphylaxis or dissemination due to the rupture of the cyst. Hence, FNAC has now been described as a safe diagnostic approach in the evaluation of suspected hydatid disease.¹⁸⁻²³ Das *et al*²⁴ reported eight cases of hydatid disease of the abdomen and thorax which were diagnosed by FNAC under ultrasound guidance. The locations of cysts were the liver in six cases, the lung in one case, and the mediastinum in one case. None of the eight cases showed any adverse reaction. Yaghan²⁵ also reported pre-operative diagnosis of mammary hydatid disease by FNAC with no anaphylactic reaction. On FNA scolices, hooklets or laminated membrane may be demonstrated. Special histochemical stains can be used for each component. The cyst membrane is stained with periodic Schiff and May-Grünwald Giemsa stain, whereas hooklets are stained with Masson's trichrome and Ziehl-Neelsen stain.

There was also a coexistent infection of *A fumigatus* along with the lung hydatid which is only occasionally seen. The diagnosis was made by fungal culture of bronchial washings and by the galactomannan test in the serum which was also positive. Mahmood *et al*²⁶ also reported a hydatid cyst complicated by an *Aspergillus* infection and the patient finally underwent surgical resection of the hydatid cyst. Coexistence of pulmonary hydatid cyst

and pulmonary tuberculosis is also seen in some cases.²⁷ The patient was treated with albendazole and voriconazole followed by lobectomy of right upper and middle lobes. Even in intact pulmonary hydatid cysts, the treatment of choice is the surgical resection of the cyst and incomplete resection is associated with high recurrence rates.²⁸ In our case of ruptured hydatid cyst, there was only one complication of secondary infection with *A fumigatus*. Ruptured hydatid cyst which persist longer than 10–14 days are predisposed to superinfection and may show increased parenchymal reaction.²⁹ Spontaneous rupture of the cyst leads to haemoptysis, cough with expectoration of the contents of the cyst and acute onset of chest pain.³⁰ Other complications of ruptured hydatid cyst include secondary echinococcosis, acute hypersensitivity reactions, pneumothorax and pleural effusion.

Learning points

- The provisional diagnosis made on radiological examination may not always be correct on investigating the patient further. In such cases the atypical presentation of other diseases should be considered in differential diagnosis especially the diseases endemic in the particular area. Here, we have highlighted an uncommon presentation of hydatid disease of the lung diagnosed by video bronchoscopy and bronchoalveolar lavage (BAL) as clinico-radiological examination along with fine-needle aspiration cytology of the lesion proved insufficient tools to make a diagnosis.

Competing interests None.

Patient consent Obtained.

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