Unusual presentation of more common disease/injury

An unusual cause of haemoptysis: a diagnostic challenge for clinicians

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Summary

A 32-year-old male patient presented with haemoptysis in the background of high-grade fever for 3 weeks. Chest examination and x-ray were suggestive of right-sided moderate pleural effusion. On finding tender hepatomegaly in abdominal examination, an ultrasonography of abdomen was performed which was suggestive of ruptured hepatic abscess. Cytological examination of both sputum and aspirate from hepatic abscess showed neutrophilic debris mixed with red blood cells. The serological test for antibody to *Entamoeba histolytica* was positive. Computerised tomography-guided trans-tracheal fistulogram demonstrated presence of hepato-bronchial fistula. Our case responded to conservative management. Follow-up ultrasonography after 6 months showed total abolition of abscess cavity and sealing of bronchial connection. Amoebic liver abscess complicating into hepato-bronchial fistula is thought to be an obsolete entity in contemporary world. But possibility of amoebic liver abscess should be kept in mind while managing a patient of haemoptysis in appropriate clinical setting in endemic areas.

BACKGROUND

Haemoptysis secondary to an abdominal pathology does not come readily in mind and can be a manifestation of pleura-pulmonary amoebiasis in developing countries including India. Although improved sanitation and antimicrobial treatment markedly reduced the occurrence of amoebic liver abscess (ALA) and its complications, ALA remains the third most common cause of casualties from parasitic diseases worldwide.¹ ALA complicating to hepato-bronchial fistula (HBF) is rare but not eradicated from the contemporary world questioning the efficacy of conventional amoebicidals against *Entamoeba histolytica* in the light of emerging antimicrobial resistance. Although surgical repair of fistula or drainage of abscess can be employed, medical management alone can produce favourable outcome. Over the years, demonstration of HBF had been challenging. In most of the earlier literature, HBF is demonstrated during autopsy or surgical exploration. Here the authors employed a novel diagnostic approach—CT-guided fistulogram following administration of non-ionic water-soluble contrast material with 4% xylocaine viscous solution diluted in 1:1.5 ratio puncturing cricothyroid membrane which was indicative of hepatobronchial fistula.

CASE PRESENTATION

A 32-year-old man, smoker and alcoholic for last 8 years, admitted to a peripheral hospital with intermittent high-grade fever for 3 weeks. He also complained of non-exertional right-sided chest pain, shortness of breath and cough with thick muco-purulent expectoration for 7 days. One week later, the patient developed recurrent bouts of haemoptysis which was persistent even after treatment with antibiotics. After 2 units blood transfusion he was referred to our hospital. The patient was non-diabetic and

there was no contact with tuberculosis. There was no history of jaundice or upper gastrointestinal bleeding in the past but significant weight loss was present over last 1 month. On general examination, raised temperature, pallor, tachycardia and tachypnoea were noted. Chest examination revealed dull percussion note and diminished breath sound in the right lower zone. Abdominal examination pointed out a soft tender hepatomegaly with a span of 18 cm with tenderness from right fifth intercostal space. The remainder of systemic examination was unremarkable.

INVESTIGATIONS

Routine blood examination revealed a total leucocyte count of 13 000/dl with 80% neutrophil and serum alkaline phosphatase was 474 U/l (normal 98–249 U/l). Chest x-ray showed elevated right dome of diaphragm with homogenous opacity involving right lower lung-field (figure 1). On aspiration, pleural fluid was straw-coloured and exudative in nature with adenine deaminase of 10 U/l (normal <30 U/l). Ultrasonography (USG) of abdomen revealed a solitary hypo-echoic lesion (83 mm×75 mm) in right lobe of liver with double echogenic lines at its postero-superior margin with the presence of echogenic collection in right subdiaphragmatic space probably due to ruptured liver abscess (figure 2). In contrast to the patient's description of sputum, we found it to be thick reddish colour and plenty of degenerated cells mixed with red blood cells were found in cytological examination. USG-guided aspiration of fluid from liver abscess also revealed thick brownish red collection similar to the sputum in appearance and microscopic examination. Examination of the stool for trophozoites of *E histolytica* was made on three consecutive days and stool was negative for amoeba in all three accessions.

Figure 1 X-ray of chest postero-anterior view showing elevated right dome of diaphragm with homogenous opacity involving right lower lung-field with obliteration of right costo-phrenic angle.

CT scan of abdomen confirmed presence of hepatic abscess (80 mm×75 mm) placed in the postero-superior region of right lobe of liver (figure 3). Then a CT-guided fistulogram was performed after injection of non-ionic water-soluble contrast media with 4% xylocaine viscous solution diluted in 1:1.5 ratio through crico-thyroid membrane puncture. Passage of contrast material was noted from bronchus to the core of hepatic abscess on attaining a right lateral decubitus. Reconstructed CT scan

Figure 2 Ultrasonography of abdomen revealed a solitary hypoechoic lesion (83 mm×75 mm) in right lobe of liver with double echogenic lines (F) at its postero-superior margin with presence of echogenic collection.

image pointed towards the presence of HBF (figure 4). The amoebic serology was tested by ab108734 *E histolytica* IgG human ELISA kit with a result of 22 NTU (nephelometric turbidity unit; Cut-off: 10 NTU, Grey zone: 9–11 NTU, Negative: <9 NTU, Positive: >11 NTU). Level of serum anti-echinococcal IgG and alpha-fetoprotein were within normal limits. Markers for Hepatitis B, C and HIV infections were negative.

DIFFERENTIAL DIAGNOSIS

History and thoracic examination misled us with a possible supra-diaphragmatic pathology including pulmonary tuberculosis and aspiration pneumonia leading to lung abscess which is common in alcoholics. On the other hand, soft tender hepatomegaly with high rise of temperature was suggestive of intra-abdominal collection of pus including sub-phrenic abscess or hepatic abscess with complication. Ruptured hydatid cyst of lung or liver with rupture into bronchus was also considered. Albeit loosely fitted with the scenario, a possibility of cavitating and necrotic hepatic neoplasms with bronchial connection could not be ruled out.

TREATMENT

Treatment was initiated with intravenous ceftriaxone 2 gm 12 hourly and metronidazole 750 mg 8 hourly and continued for 14 days. Thoracic surgeons opined against surgical repair in the acute stage due to lytic action of the protozoa and advised to continue conservative management as rupture of ALA to bronchus helps in spontaneous drainage of pus and there are previous case reports where patients of HBF improved on medical therapy alone without surgical intervention.

OUTCOME AND FOLLOW-UP

Week-long antimicrobial therapy curbed the fever and cough was significantly diminished with minimal residual expectoration. The patient gradually put on weight and regained normal daily activities. A repeat USG done after 2 weeks of conservative therapy revealed diminution in the size (43 mm×35 mm) of ALA with sealing of HBF by collapsed lower zone of the right lung. The patient was discharged with oral tinidazole 600 mg three times daily (TDS) and diloxanide furoate 500 mg TDS for 14 days. Follow-up USG after 6 months demonstrated complete radiological resolution of hepatic abscess.

DISCUSSION

ALA is the most common extraintestinal manifestation of *E histolytica*. A total of 10% population all over the world are infected with *E histolytica* whereas only 10% of them are symptomatic.¹ Although a combination of high rise of temperature, right-upper-quadrant pain and substantial intercostals tenderness along with a travel to or residence in an endemic area is conventionally described with ALA, the classical picture is frequently missing.² Less than 30–40% of patients with amoebic liver abscess have concomitant intestinal amoebiasis. Hence, the microscopic examination of the stool for the identification of cysts is of little value. The diagnosis of ALA relies on radiological identification of space occupying lesion of the liver and demonstration of anti-amoebic antibody by means of indirect haemagglutination or enzyme linked immunosorbent

Figure 3 Contrast-enhanced CT of chest and upper part of abdomen before (right) and few minutes after injection of non-ionic water-soluble contrast media with 4% xylocaine viscous diluted in 1 : 1.5 ratio through crico-thyroid membrane puncture (left). Passage of contrast material was noted to the core of hepatic abscess on attaining a right lateral decubitus.

assay (ELISA). Complications of ALA include rupture into pleural, bronchial, pericardial or peritoneal spaces, obstructive jaundice and inferior vena caval thrombosis.³

Actual incidence of intrathoracic manifestations of ALA in the recent time is not known, owing to multiple factors including under-reporting but few earlier studies revealed incidences of thoracic complications can be as high as 86% in patients of ALA.^{4–5} Pleura-pulmonary diseases secondary to ALA consist of (1) reactive inflammatory response involving pleura or lung parenchyma resulting in pleural effusion or alveolar reaction producing dyspnoea; (2) rupture into pleural space resulting in empyema thoracis producing toxic feature, cough and chest pain; (3) rupture into bronchial tree with development of HBF resulting in expectoration of 'anchovy-sauce' pus, lung abscess or collapse.⁶ The classical 'anchovy sauce' appearance of sputum has been overemphasised in the past. Neither is anchovy sauce-like pus always amoebic nor amoebic pus always like anchovy sauce.

Kubitschek *et al*⁶ published a series of seven cases who were hospitalised with predominantly pulmonary symptoms and were found to have amoebic liver abscess. Lyche *et al*⁷ found that among the patients of ALA with predominant abdominal symptoms, 60% patients had pulmonary symptoms and 47% had pulmonary signs. Ibarra-Perez⁸ observed that one in three cases of pleura-pulmonary amoebic disease have an exudative reactive pleural effusion which was also present in our case as well.

Figure 4 Reconstructed CT image demonstrates the presence of hepato-bronchial fistula (yellow arrow).

Earlier studies demonstrated HBF either intraoperatively or during autopsy. In our case HBF is confirmed with the help of dual-slide CT scan which demonstrated the passage of non-ionic water-soluble contrast material into hepatic abscess following puncture through crico-thyroid membrane. Multidetector slide CT scan or MR with ultra-fast, breath-hold, heavily T2-weighted sequences could have been useful for more non-invasive depiction of HBF, if available.⁹ Historically, repair of the fistulous tract or surgical or percutaneous drainage of the abscess had been employed.¹⁰ Evidence support that antimicrobials can be used as initial treatment of HBF when detected early as it prevents further progression of intra-thoracic complications and decreases need for surgical intervention.^{4–8} Spontaneous sealing of fistulous tract along with shrinkage of abscess cavity was noted within 3 weeks of conservative management and in 6 months complete radiological resolution of hepatic abscess was achieved in our case.

Learning points

- ▶ Amoebic liver abscess complicating into hepato-bronchial fistula is not an extinct entity from contemporary world and can be rarely found in developing countries like India.
- ▶ Recognition of intrathoracic amoebiasis is essential among residents of endemic areas and also among immigrants elsewhere suffering from unexplained respiratory problems.
- ▶ Early treatment of amoebic liver abscess complicating to hepato-bronchial fistula with amoebicidal alone prevents the progression of intrathoracic complications and decreases the need for surgical intervention.

Acknowledgements The authors would like to acknowledge Dr Anup Sadhu, Consultant Radiologist for radiological insights and Ms Corie Goodloe of Washington University for her assistance with writing this case report.

Competing interests None.

Patient consent Obtained.

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Please cite this article as follows (you will need to access the article online to obtain the date of publication).

Talukdar A, Mukherjee K, Khanra D, Saha M. An unusual cause of haemoptysis: a diagnostic challenge for clinicians. *BMJ Case Reports* 2012;10.1136/bcr-2012-006751, Published XXX

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