Case Report

An unusual cause of haemoptysis: hepatobronchial fistula

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ABSTRACT

Haemoptysis is a common clinical problem but a non-specific symptom and can occur in about 100 different clinical conditions. Diagnosing aetiology is not always straight forward. Authors present a case of Hepatobronchial fistula (HBF) which initially diagnosed as tuberculosis due to its atypical presentation and rarity of HBF.

Keywords: Haemoptysis, Hepatobronchial fistula, Liver abscess, Pulmonary amoebiasis

INTRODUCTION

Hemoptysis is defined as the expectoration of blood from the lung parenchyma or airways. Haemoptysis is a common clinical problem reported to be the cause of attendance in 7-15% of patients coming to chest clinics.1 However, it is a non-specific symptom and can occur in about 100 different clinical conditions.2 Expectoration of even relatively small amount of blood is an alarming symptom and can be a marker for potentially dangerous disease like bronchogenic carcinoma. On the other hand, massive haemoptysis can represent an acutely life threatening problem. Therefore, haemoptysis of any degree needs thorough evaluation. The aim of evaluation is to find treatable cause and at times, to reassure the patient. Although in some cases the aetiology may be readily apparent, in others it may be challenge for clinician. Even after giving best of clinician efforts, up to 30% of patients show no identifiable aetiology for their haemoptysis.3 Common causes of haemoptysis in developing country are pulmonary tuberculosis, bronchitis, bronchiectasis, lung abscess, pulmonary embolism, and infarction. Less common causes are renal, cardiac, collagen vascular diseases, etc. Authors came across a case who was clinically having all symptom suggestive of pulmonary tuberculosis. In county like India where incidence of pulmonary tuberculosis is high and considered in all differential diagnosis until unless proved otherwise. This case was unique so challenging also because patient was having symptoms all related to respiratory system but lesion was in abdomen. Authors are here presenting a case of Hepatobronchial fistula, a rare cause of haemoptysis in the modern era of antibiotic. Patient was not having risk factor for hepatobronchial fistula Hepatobronchial fistulae (HBF) are rare entities. They are defined as abnormal communications of a sector of liver parenchyma with a sector of the bronchial tree through a diaphragmatic pathway. First described by Peacock in 1850 in a patient with a hepatic hydatid cyst and hydatid vomica, its frequency has decreased, mainly due to the use of antibiotics in the presence of hepatic abscesses and the surgical treatment of hepatic and pulmonary hydatid cysts.4 HBF may be congenital or acquired. Acquired HBF (80%) is mainly caused by hepatic hydatid cysts that migrate toward the pleural cavity through the diaphragm. The other 20% are due to hepatic abscesses (amebic or pyogeneric), lithiasis of the biliary tract and, less frequently, as a result of surgery or liver trauma.5,6

CASE REPORT

A 34-year old male presented with history of low grade intermittent fever for six weeks duration followed by coughing up blood for four week duration. It was associated with anorexia, general malaise, night sweats
and significant weight loss. There was no history of breathing difficulty, chest pain, urinary or bowel complaints or yellow discoloration. He was an otherwise healthy heterosexual male, without drug use or high risk behavior.

The patient was started on antituberculous treatment (ATT) in a local hospital but he was not having improvement continuing ATT for one month. Physical examination revealed that the patient was having fever (max 100 f) blood pressure 110/70 mmHg, heart rate was 86 beats/min a respiratory rate of 20 breaths per minute.

Oxygen (O2) saturation was 100% on room air. The patient was alert, and appropriate with no signs of respiratory distress. More concerning was crepitation on the right basal side of the chest on auscultation. His abdomen was having tenderness over right (Rt) upper quadrant. Abdomen was soft, bowel sounds were present. The remainder of the physical examination was non-contributory.

Laboratory reports were as follows; blood haemoglobin 10g/dl; urea- 24mg/dl; creatinine- 0.7mg/dl; total counts-8500cells/mm³, Differential counts: Neutrophil 72%, Lymphocyte- 24%; Eosinophils 4%. Erythrocyte sedimentation rate (ESR) was 8mm in 30 minutes and 18mm in one hour. Sputum AFB (acid fast bacilli)-negative. Markers for Hepatitis B, C and HIV infections were negative. Stool examination-no evidence of parasites. Serum: positive for Entamoeba histolytica antibody (latex agglutination test). Chest x-rays did not contribute any pathology (Figure 1).

Ultrasonography (USG) of abdomen (Figure 2, 3 and 4) revealed a solitary hypo-echoic lesion in right lobe liver. There was also a hypo-echoic tract extending from the sub diaphragmatic collection piercing the diaphragm communicating with Rt lower lobe representing hepatobronchial fistula.

In contrast to the patient’s description of sputum, authors found it to be thick reddish color 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.

CECT abdomen (Figure 5) confirmed an ill-defined hypo dense lesion (measuring 9.2x 6.0 cm) in Right lobe of liver in sub capsular region with minimal adjacent sub hepatic collection.
The largest burden of E. histolytica infection is seen in Central and South America, Africa, and the Indian subcontinent. Amoebic infection caused by the protozoan parasite Entamoeba histolytica is a widespread endemic infection in many of the tropical and subtropical areas of the world. Amoebic liver abscesses occur most commonly in the age group of 20–45 years and have been noted infrequently in the extremes of ages, with an adult male to female ratio of 10:1.10

The diagnosis of amoebic liver abscess is sometimes difficult and delayed since its clinical manifestations are highly variable, like in our patient who is 34 years male presented with fever and haemoptysis for the past four to six weeks, in absence of symptoms like right upper quadrant abdominal pain, or jaundice.

Authors report a case of Hepatobronchial fistula secondary to an amoebic liver abscess that was misdiagnosed as tuberculosis. Pleuro-pulmonary complication of amoebiasis is easily confused with other illnesses, and it is treated as pulmonary TB, bacterial lung abscess, and carcinoma of the lung.11

Amebic infection is either intestinal, producing amebic dysentery, or extraintestinal, producing mainly amebic hepatitis. Entamoeba histolytica gains access to the colon following ingestion of food or drink contaminated with the cysts of the parasite.

Entamoeba histolytica, an amoebic protozoan parasite, is the most invasive of the Entamoeba group. In the stomach the wall of the cysts is digested, liberating the parasite, which through its cytolytic enzymes finds its way to the submucosa of the colon, where it produces the characteristic flask-shaped ulcers. At any time during this phase of amebic colitis, Entamoeba histolytica may erode branches of the portal vein and reach the liver as emboli in the portal blood. Once established in the liver sinusoids, the amebae produce cytolytic destruction in the liver. This process may be diffuse, giving rise to diffuse amebic hepatitis, or localized, producing an amebic liver abscess, which is commonly found in the right lobe of the liver and less often in the left lobe. Characteristically, the abscess is solitary, containing chocolate brown or anchovy-colored saucelike pus that is sterile on culture.

Amebic liver abscess may be secondarily infected; it may rupture in the sub-pancreatic area or general peritoneal cavity; it may erode the diaphragm leads to Thoracic Empyema or rupture into bronchus with the expectoration of Anchovy-paste coloured pus from amoebic liver abscess.

Pulmonary amoebiasis, the second most common extraintestinal pattern of infection, is frequently associated with amebic liver abscesses. It occurs in 2-3% of patients with invasive amoebiasis.7,8 Lung disease without liver involvement is exceptional and it is believed...
that infection of the lung is a result of haematogenous spread from a primary site, usually colon.8

Typical symptoms include abdominal pain and fever. Abdominal pain is reported to be present in 98% and fever in 74% of the cases. A history of diarrhoea is present in 20-30% of the cases. Hepatomegaly and tenderness in the right upper quadrant of the abdomen (over the liver) are the most frequent physical signs.

This patient was not having abdominal pain, may be due to being decompressed through communicating bronchus regularly. In this situation diagnosing liver abscess clinically became more difficult until unless you have high index of suspicion. This patient was coughing up characteristic amoebic pus which is a pathognomonic sign of hepatobronchial fistula.

Diagnosis of Hepatobronchus fistula was based on positive serum antibodies to amoeba, imaging of solitary liver abscess draining into bronchus. Diagnostic serological test for antibody against Entamoeba histolytica done by Latex agglutination method is rapid, having sensitivity and specificity of 98 and 96% respectively.

Management of HBF is multi steps procedure depends upon complication, basically consist of draining of pus, antibiotic treatment and surgical repair of permanent damage. This case was managed satisfactorily with course of antibiotic only as pus was already being drained through bronchus and fistula got closed later on by collapsed lung. There are previous case reports where patients of HBF improved on medical therapy alone without surgical intervention.

CONCLUSION

Cause of haemoptysis is not always straight forward, lesion may be located outside of chest and diagnosis may be delayed due to an unusual presentation. High index of suspicion is therefore required to diagnose. Complication is not bad all the time as rupture of liver abscess to bronchus helps in spontaneous drainage of pus.

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REFERENCES

1. Khadra I, Braun SR. Haemoptysis In: Braun SR. Concise textbook of Pulmonary Medicine 1st ed. Elsevier Science publishing Co., New York; 1989:903-608.
2. American Thoracic Society. The management of haemoptysis. Am Rev Respir Dis. 1966:93:471-4.
3. Weinberger SE, Braunwald E. Cough and Haemoptysis In: Braunwald E, Fauzi AS, Kasper DL, Hauser SL, Longo DL, Jameson JL. (Eds) Harrison’s Principles of Internal Medicine 15th ed, McGraw Hill; 2001:203-207.
4. Peacock TB. Case in which hydatids were expectorated and one of suppuration of hydatid cyst of the liver communicating with the lungs. Edinburgh Med J. 1850 Jul 1;74(184):33-46.
5. Eryigit H, Oztas S, Urek S, Olgac G, Kurutepe M, Kutlu CA. Management of acquired bronchobilary fistula: 3 case reports and a literature review. J Cardiothor Surg. 2007 Dec 1;2(1):52.
6. Talukdar A, Mukherjee K, Khanra D, Saha M. An unusual cause of haemoptysis: a diagnostic challenge for clinicians. BMJ Case Rep. 2012;2012.
7. Neghina R, Neghina AM, Merkler C, Marinescu I, Iacobiciu I. A case report of pulmonary amoebiasis with Entamoeba histolytica diagnosed in western Romania. J Infect Developing Countries. 2008;2:400-2.
8. Gupta KB, Manchanda M, Chaudhary U, Verma M. Superior Vena cava syndrome caused by pulmonary amoebic abscess. Indian J Chest Dis Allied Sci. 2006;48:275-7.
9. Singh U, Petri Jr WA. Amebas. In: Gillespie SH, Pearson RD, editors. Principles and practice of Clinical Parasitology. Chichester: John Wiley and Sons, Ltd. 2001: 197-218.
10. Karkhanis VS, Joshi JM. Pleural effusion: diagnosis, treatment, and management. Open Access Emerg Med. 2012;4:31-52.
11. Hidron A, Vogenthaler N, Santos-Preciado JI. Cardiac involvement with parasitic infections. Clin Microbiol Rev. 2010: 23:324-49.

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