International Journal of Research in Medical Sciences
Suman S. Int J Res Med Sci. 2020 Aug;8(8):3102-3105
www.msjonline.org
pISSN 2320-6071 | eISSN 2320-6012
DOI: http://dx.doi.org/10.18203/2320-6012.ijrms20203473

Case Report

An unusual cause of haemoptysis: hepatobronchial fistula

Sonu Suman*

Department of Medicine, Military Hospital, Saugor, Madhya Pradesh, India

Received: 03 June 2020
Accepted: 02 July 2020

***Correspondence:**
Dr. Sonu Suman,
E-mail: 15680h@gmail.com

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: Heamoptysis, Hepatobronchial fistula, Liver abscess, Pulmonary amoebesis

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, upto 30% of patients show no identifiable aetiology for their haemoptysis.[3] Common causes of haemoptysis in developing countary 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 countary like india where incidence of pumonary tuberculosis is high and considered in all differential diagnosis until unless proved otherwise. This case was unique so challanging 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 heamoptysis 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 pyogenic), 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 continueing ATT for one month. Physical examination revealed that the patient was having fever (max 100 f) blood pressur 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 xrays did not contribute any pathology (Figure 1).

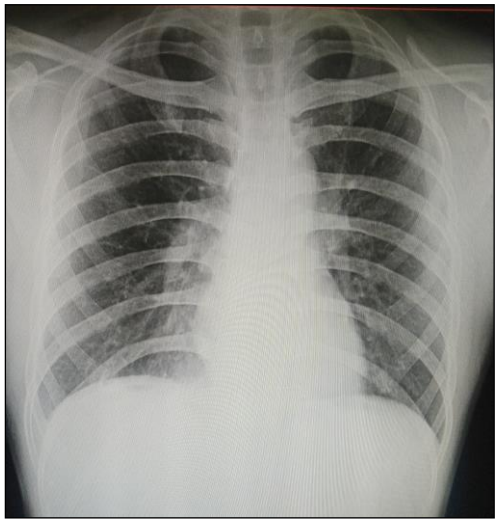

Figure 1: Normal chest x rays.

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.

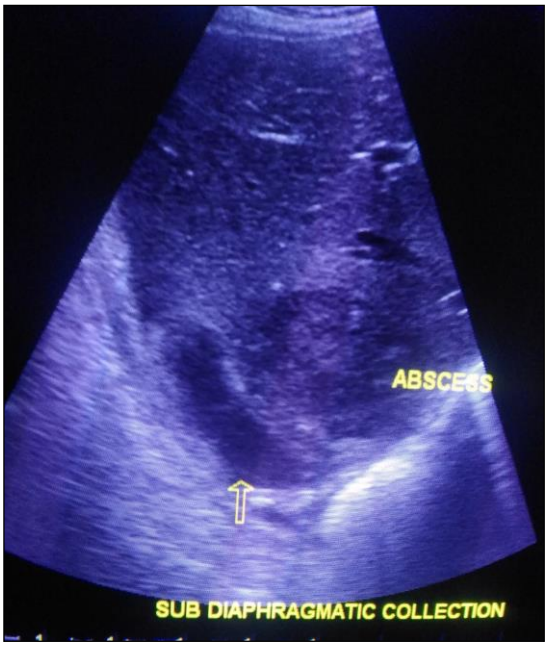

Figure 2: Liver abscess and sub diaphragmatic collection.

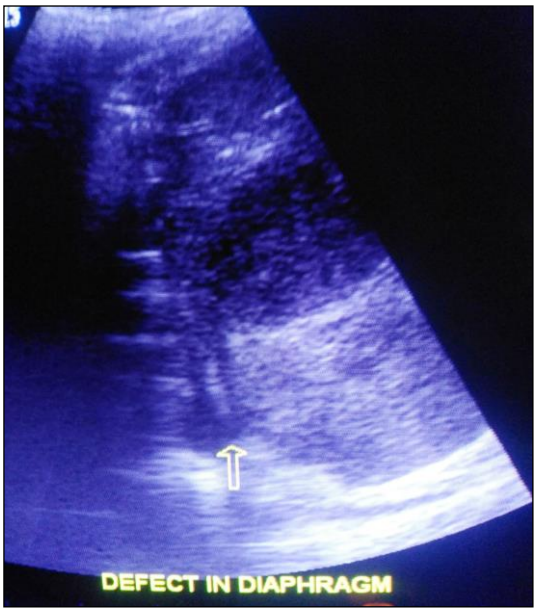

Figure 3: Defect in diaphragm.

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.

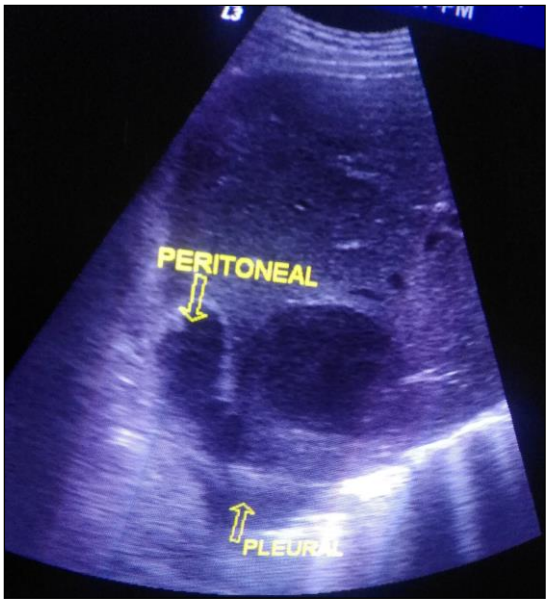

Figure 4: Liver abscess with fistula connecting liver to lung.

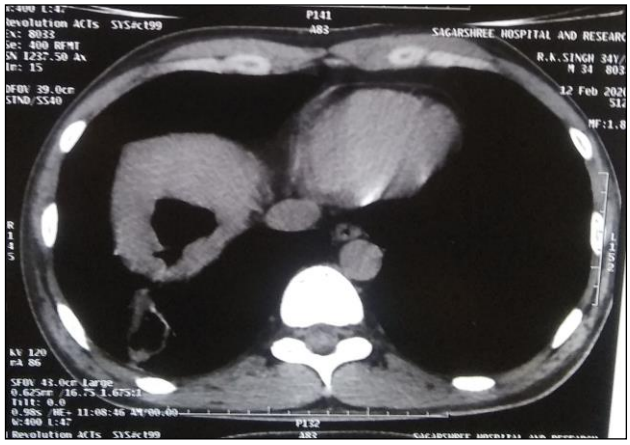

Figure 5: Hypodense lesion in Rt lobe.

Patient was treated with intravenous ceftriaxone 2 gm 12 hourly and metronidazole 750 mg 8 hourly and advised to continue conservative management for 14 days as rupture of abscess 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 or percutaneous drainage.

Patient gradually improved. Fever subsided by seven day and follow up USG after two weeks confirmed diminution in the size of liver abscess with sealing of HBF by collapsed lower zone of the right lung.

DISCUSSION

Among parasitic diseases, amebiasis caused by Entamoeba histolytica, is the third most frequent cause of mortality after malaria and schistosomiasis.[7] Developing countries are the most affected by this disease.[8] The largest burden of E. histolytica infection is seen in Central and South America,Africa, and the Indian subcontinent.[9] Amebic 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.[10,11]

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-phrenic area or general peritoneal cavity; it may erode the diaphragm leads to Thoracic Empyma or rupture into bronchus with the expectoration of Anchovy-paste coloured pus from amoebic liver abscess.

Pulmonary amebiasis, the second most common extraintestinal pattern of infection, is frequently associated with amebic liver abscesses. It occurs in 2-3% of patients with invasive amebiasis.[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 situaltion diagnosing liver abscess clinically became more difficult until unless you have high index of suspicion. This patient was coughing up characteristic amebic 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 agglutation mathod 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 satisfactory with course of antibiotic only as pus was already being drained through bronchus and fistula got closed later on by collaped 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.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Suman S. An unusual cause of haemoptysis: hepatobronchial fistula. Int J Res Med Sci 2020;8:3102-5.