Case Report

Brocho-biliary fistula: A rare complication after ruptured liver abscess in a 3½ year old child

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ABSTRACT

Bronchobiliary fistula (BBF) is a rare condition, defined by the presence of abnormal communication between biliary tract and bronchial tree. We describe a 3½-year-old child who developed BBF after rupture of liver abscess. She underwent exploratory laparotomy and peritoneal wash for ruptured liver abscess. Seven months later she presented with fever and cough with yellow-colored expectoration (bilioptysis). An abnormal communication between right branch of the hepatic duct and a branch of right main bronchus was identified. Child underwent right lateral thoracotomy and right lower lobectomy with surgical excision of sinus tract. On follow-up child was asymptomatic and doing well.

KEY WORDS: Bilioptysis, bronchobiliary fistula, liver abscess

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INTRODUCTION

Bronchobiliary fistula is an abnormal communication between biliary tract and bronchial tree. It is a rare condition in children with only few cases been reported in literature.[1-5] It may be congenital or acquired post-abdominal surgery or trauma. We report a case of bronchobiliary fistula which occurred as a complication of ruptured liver abscess and this complication has not been reported earlier.

CASE REPORT

A 3½-year-old girl presented with complaints of fever, cough and respiratory distress with expectoration of yellowish sputum since past 12 days. Past history revealed that she had an episode of high-grade fever with yellowish discoloration of conjunctiva, pain abdomen and generalized swelling of body with altered sensorium 6 months back. For these complaints, she had then been evaluated and CT abdomen had shown ruptured abscess of right lobe of liver, for which she had underwent exploratory laparotomy, drainage of liver abscess and peritoneal wash. Peritoneal, pleural and abscess drains had been sequentially removed post-operatively. She had also undergone a second laparotomy and adhesiolysis, 40 days following the first surgery for intestinal obstruction. Subsequently she remained asymptomatic for next 2 months and then started developing fever, persistent cough with yellowish expectoration and difficulty in breathing.

At admission she was febrile with respiratory distress (nasal flaring and subcostal/intercostal retractions were present) and oxygen saturation in room air was 89%. Her blood pressure and capillary refill time were within normal limits. Chest examination revealed decreased air entry on right side and bilateral scattered crepitation (R > L) and conducted sounds. Per abdomen examination revealed dilated superficial veins and scar mark of previous laparotomy. Liver was palpable 1.5 cm below costal margin with span of 7.5 cm, and spleen was just palpable. Cardiovascular and central nervous system examination were normal. Her height and weight was less than 3rd percentile according to WHO growth charts.

Investigations revealed hemoglobin of 10.4 gm/dL, total leukocyte count of 11,700/mm^3 with 69% neutrophils, 30% lymphocytes, 1% eosinophils and platelet count of 4.1×10^5 per microliter. Peripheral blood smear examination was normal. Mantoux test was negative. Gastric aspirate for acid-fast bacilli (AFB) was negative on two occasions. The sputum smears and cultures were negative for bacteria and AFB. Human immunodeficiency virus (HIV) serology was

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also negative. Other immunodeficiency work-up including CD4/CD8 counts, nitroblue tetrazolium tests (NBT) test for chronic granulomatous disease and immunoglobulin profile were within normal limits. Liver function tests were within normal limits. Chest X-ray showed homogenous opacity of right lower zone with blunting of right costophrenic angle [Figure 1a]. Ultrasound abdomen was done which showed altered architecture of right lobe of liver with minimal right pleural effusion. Doppler imaging showed attenuation of right branch of portal vein.

Bronchoscopy revealed yellowish frothy secretion coming out of right main bronchus. A possibility of bronchobiliary fistula was kept, and broncho-alveolar lavage (BAL) fluid was sent for relevant investigations. BAL fluid revealed bilirubin of 2.5 mg/dL with conjugated fraction of 2 mg/dL. CECT chest and abdomen revealed trans-diaphragmatic bronchobiliary fistula with resolving abscess in right lobe of liver and consolidation of right lower lobe of lung [Figure 1b and c]. Magnetic resonance cholangiopancreatography (MRCP) revealed abnormal tract communicating between a tributary of right branch of the hepatic duct and a branch of right lower lobe bronchus [Figure 1d]. HIDA scan revealed good hepatocyte function, bronchobiliary fistula with extension of tracer in to right bronchus, trachea and abnormal tracer accumulation in right lower lobe of lung with patent bilio-enteric pathway [Figure 2]. Child underwent right lateral thoracotomy and right lower lobectomy with surgical excision of sinus tract under general anesthesia. Chest drain was removed on post-operative day 5. Child remained asymptomatic and was discharged on post-op day 10. On follow up after 3 months, she remained asymptomatic with no respiratory distress and was gaining weight.

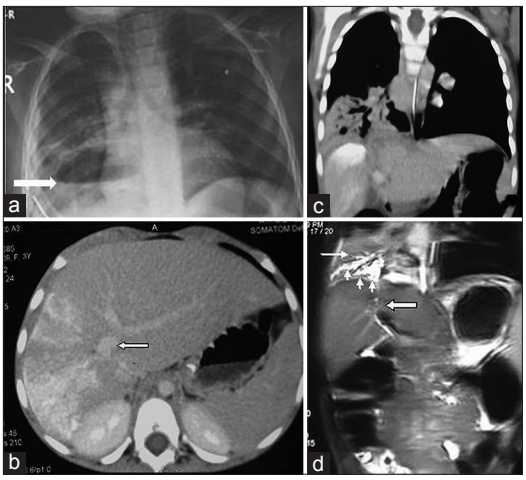

Figure 1: (a) Chest radiograph PA view shows right-sided pleural effusion and a large air-fluid level at right lower hemithorax (block arrow) (b) Axial CECT image of liver reveals reduced volume of the right lobe; associated with diffuse hyperdense attenuation of the entire lobe. Note absent opacification of the right branch of portal vein whereas left branch is well visualized (arrow) (c) Coronal reformatted image of CECT of chest and abdomen reveals extensive right lower lobe consolidation with areas of cavitation (d) MRCP shows a hyperintense track containing fluid could be traced (block arrow); extending from the liver (right hepatic duct) to the lower lobe bronchi, suggesting a broncho-biliary fistula

DISCUSSION

We report a young girl who developed bronchobiliary fistula as a complication of ruptured liver abscess and was treated with surgery. BBF may be congenital; however, in most cases, it is acquired and parasitic infection of liver (Hydatid cyst) is the commonest cause.[6,7] Other causes of BBF include trauma, sub-diaphragmatic abscess, post-surgical states, and lithiasis in biliary trees, cholecystitis/pancreatitis, and liver/biliary tree tumor and radiofrequency ablation.[8]

BBF is extremely rare in children. Although the majority of BBF cases have been reported in neonates, age of presentation may vary from infants to adults. Only a few cases have been reported in the literature.[1-5] BBF occurring as a complication of ruptured liver abscess in children has not been reported yet.

The clinical presentation is predominately pulmonary, with abdominal symptoms being less frequent. An irritating cough and expectoration of yellowish sputum termed as 'bilioptysis' is the typical presentation of BBF. Sutherland *et al*.[5] have described false bile ptyalism (excessive salivation) in patients with sickle cell disease and hemolytic crisis. However, if this condition can be excluded, the presence of bile in the sputum is pathognomonic of bronchobiliary fistula. Our case presented with recurrent fever, chronic cough following treatment of ruptured liver abscess. She developed yellowish, bile-stained sputum (bilioptysis) suggesting formation of a track from liver to pleural cavity through diaphragm and lately developed bronchobiliary fistula.

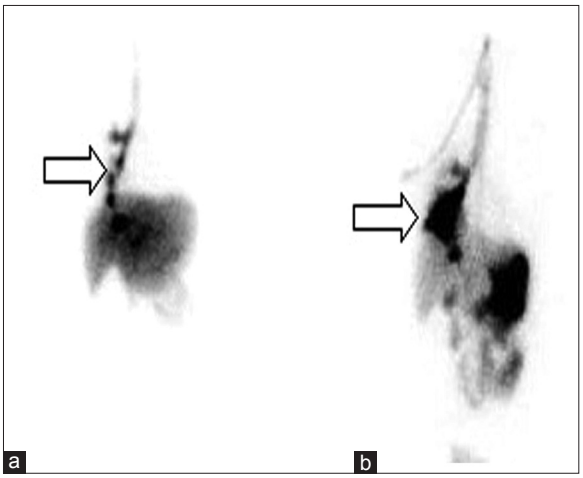

Figure 2: (a) HIDA scan reveals reflux of the tracer agent through an abnormal communication in right bronchus (b) Abnormal tracer agent also accumulated in the right lower lobe of lung with patent bilio-enteric pathway

BBF can be treated endoscopically[9] or surgically. In the acute fulminating form, an aggressive and rapid approach is necessary. A two-stage approach can be used: (1) external biliary drainage by percutaneous or surgical drainage of subphrenic abscess and/or direct percutaneous drainage of the intrahepatic biliary tract; and (2) treatment of the underlying cause. Persistent fistula with a patent biliary channel is an indication for thoracotomy. A delay in surgery might result in further damage of the lung.[10] In our patient the right lower lobe of lung was completely damaged and chronic track had formed from liver to pleura. Therefore, it was decided to perform right lower lobectomy and repair of diaphragm.

CONCLUSIONS

We conclude that bronchobiliary fistula is rare in children; a high index of suspicion is required for early diagnosis. An irritating cough with yellowish expectoration might indicate bronchobiliary fistula.

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