Cholethorax Secondary to Pleurobiliary Fistula (PBF) Post-Hydatid Cyst Surgery

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Declaration of conflicting interests

The authors declare that there is no conflict of interest.

Abstract

Cholethorax is a rare complication of hepatic hydatid disease and hepatic surgery. We report a case of Cholethorax, post hydatid cystectomy in a 28 years old male who was referred to our unit with worsening shortness of breath, cough and persistent high-grade fever. He had right sided pleural effusion with raised pleural fluid bilirubin levels, diagnosed as Cholethorax, initially managed by tube thoracostomy, endoscopic biliary stenting and then later on decortication with diaphragm repair. We believe that Cholethorax can be developed as a delayed complication of hepatic hydatidosis, as in our case who presented 2 months after surgery.

Key words : Cholethorax; Hydatid Cyst; Pleural effusion; Pleuro-biliary fistula

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Introduction

Pleuro biliary fistula (PBF) leading to Cholethorax is a rare presentation. Causes of PBF can be broadly divided into three categories: infections (echinococcosis, amoebic, bacterial), sub-hepatic or sub-phrenic abscesses and iatrogenic secondary to biliary tree manipulation. PBF, usually, is underdiagnosed condition. We report a case of 28 years old male who developed PBF,2 months after surgery for hydatid disease of the liver.

Case Report

A 28 years old young male with history of surgery for hydatid disease of the liver presented to our unit with worsening shortness of breath, high grade fever and cough. He had undergone surgery for hepatic ecchinococus infection 2 months back. On examination, he had a temperature of 40 degree Celsius, respiratory rate of 31 bpm, heart rate of 105 bpm. He was ill looking and had reduced chest movements on the right side with stony dull percussion notes and reduced air entry. No abnormal findings were observed on the left side of the chest. Rest of the systemic examination was normal. He was started on anti-tubercular therapy (ATT) by a GP for his pleural effusion, based on the finding of X-ray chest.

Investigations: Chest radiograph followed by ultrasound confirmed right sided pleural effusion. Pleural tap was done and greenish color fluid was sent for routine examination i.e. PH, Sugar, proteins, cells with differential count, gram and ZN stain. Due to the greenish yellow color of fluid and prior history of hepatobiliary surgery, pleural fluid bilirubin levels were also done. Pleural tissue was taken for histopathology through Abrams needle and fluid samples sent for C/S. CBC showed neutrophilic-leukocytosis. Rest of blood investigations were within normal limits.

CT thorax and abdomen revealed right pyopneumothorax, right lower lobe consolidation,

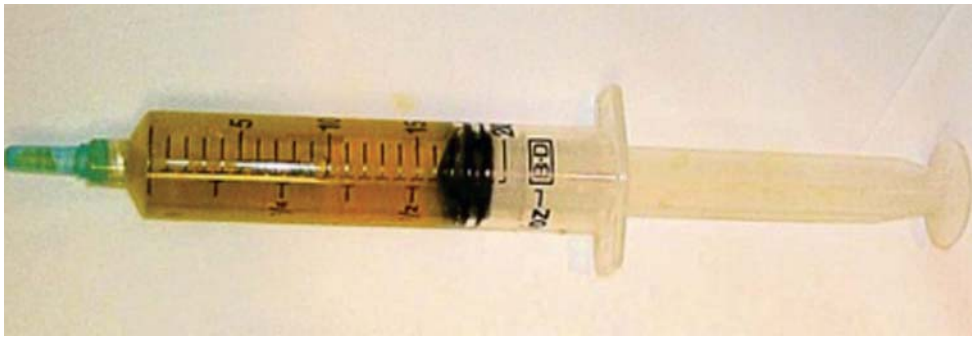

Figure 1: Appearance of Pleural fluid

fibrocystic changes, Pleural thickening and a hypodense collection in segment VII of right hepatic lobe measuring 5.3 * 3 cm with enhancing walls.

ERCP was done and plastic stent was place to ensure smooth flow of bile. Common bile duct was normal. HIDA scan showed an abnormal collection of biliary tracer in postero-superior portion of right lobe of liver

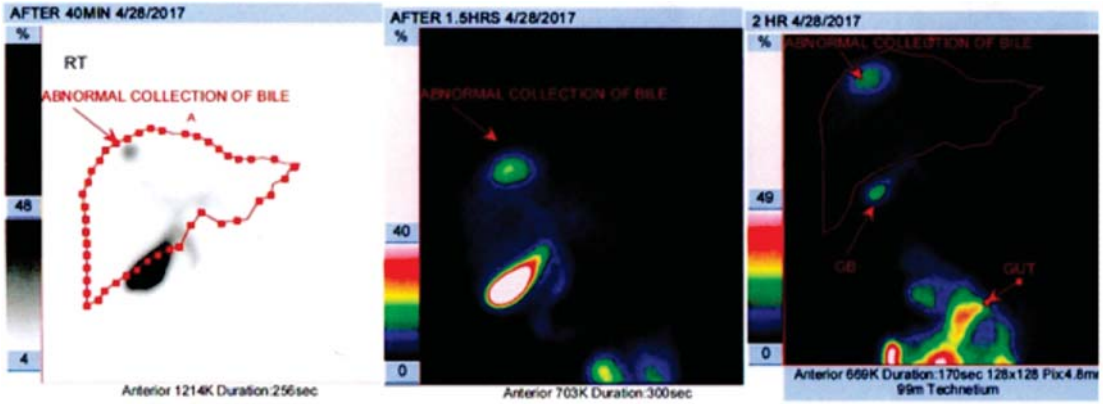

Figure 2: HIDA Scan Management:

with small and confined leak. There was smooth flow of bile through a patent plastic stent.

Management

After aspiration of turbid greenish colored fluid, a provisional diagnosis of empyema was made and right sided chest tube thoracostomy was performed which drained almost 2 L of fluid in first 2 days with subsequent 800 to 1000 ml per day. The fluid bilirubin levels were 3.63g/dl with a ratio of fluid to serum bilirubin of more than 9. The patient was diagnosed as a case of pyocholethorax and Anti tubercular therapy was stopped. Fluid C/S showed growth of pseudomonas which was sensitive to Ciprofloxacin, ceftazidime, Meropenem, amikacin. Started on sensitive antibiotics (Ceftazidime plus Amikacin). Endoscopic retrograde cholangiopancreatography(ERCP) was performed with sphincterotomy and a 10Fr, 10cm plastic stent was placed in CBD to ensure smooth flow of bile to the intestine. Hepatobiliary iminodiacetic acid (HIDA) scan showed normal flow to the gut, patent plastic stent and an abnormal tracer uptake in postero-superior portion of right hepatic lobe with confined leak (Figure_2). The drainage decreased to 100 ml per day and fluid became clearer. Due to un-expanded lung and pleural thickening the case was discussed in multidisciplinary team meeting and it was decided to go for decortication.

Open thoracotomy was performed on 20th day of admission. Broncho pleural fistula and rent in diaphragm were repaired. Thick peel over right lung was also removed. Chest tube was removed on 10th post-operative day with full expansion of the lung. The patient was discharged in stable condition.

Follow up

Full expansion of right lung was achieved after decortication. Plastic stent removed after 8 weeks on an outpatient basis. Patient is stable after 5 months from stent removal.

Discussion

Cholethorax is a rare and atypical presentation.[1] Diagnosed as demonstration of biliary fluid in pleural space. The ratio of fluid to serum bilirubin greater than 1 and pleural glycolic acid are important diagnostic markers.[2]

Most cases reported till date support surgery or procedures on biliary treeand hepatic trauma as important causative factors. Other causes of Pleuro-biliary fistula are hepatic hydatid disease, hepatic amebiasis, gallbladder diseases, haptic abscesses, malignancies, radiation and idiopathic.[3] In our case, the cause was most probably hepatic hydatid disease and the subsequent surgical removal of the hydatid cyst.

Cholethorax is a dangerous clinical entity. Due to the corrosive nature, bile causes extensive local fibrosis and pleural thickening. In some cases, ARDS may develop as a consequence of bile in the pleural cavity. In this patient, the late presentation and extensive pleural thickening induced by bile, may have led to failure of the affected lung to expand completely following tube thoracostomy and necessitated the need for decortication.

Our case is unique in that the patient developed bilothorax very late i.e. almost 2-month post-surgery. Moreover, due to high prevalence of ecchinococus infections in this part of the world require extreme vigilance and proper approach to rule out presence of bile in pleural cavity.[4]

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