Case Report

A rare pleural effusion in a young male

Shabana Begum, Subhasis Mukherjee[1], Debabani Biswas[1], Amartya Kumar Misra[1], Priyanka Ghosh[1], Pulakesh Bhanja[1]

Department of Anatomy, North Bengal Medical College and Hospital, Sushrutanagar, Siliguri, Darjeeling, [1]Department of Pulmonary Medicine, College of Medicine, and Sagar Dutta Medical Hospital, Kolkata, West Bengal, India

ABSTRACT

A 28-year-old male presented with fever with right-sided chest pain for 2 weeks. Clinicoradiological picture was suggestive of right-sided pleural effusion. He had history of polytrauma following a road traffic accident and had to undergo emergency laparotomy a month ago. Microscopic and culture examination of the pleural fluid showed neutrophilia, high bilirubin content and presence of gram-negative bacilli. Ultrasound of the abdomen showed the presence of biloma in the liver and right subdiaphragmatic space with fistulous communication into the right thoracic cavity. The patient was managed successfully with complete recovery.

KEY WORDS: Biliopleural fistula, biloma, pleural effusion

Address for correspondence: Dr. Subhasis Mukherjee, 181/2B, Roypur Road, Kolkata - 700 047, West Bengal, India. E-mail: drsubhasismukherjee@yahoo.in

INTRODUCTION

Bilious pleural effusion is an extremely rare cause of pleural effusion.[1,2] It usually occurs after surgical exploration of the biliary tree,[3,4] but traumatic disruption of hepatobiliary system leading to bilothorax is also reported in the literature.[2] Irrespective of the etiology, thoraco-biliary fistulous and bilothorax put the clinicians in diagnostic as well as therapeutic dilemma. We report a case of delayed formation of biloma following a road traffic accident (RTA) complicated with a subdiaphragmatico-pleural fistula, a month after the RTA.

CASE REPORT

A 28-year-old male, auto-rickshaw driver by profession, presented to us with high grade fever and right-sided pleuritic chest pain for a duration of 10 days. He had a history of RTA about 6 weeks ago. He sustained a blunt injury to his abdomen and a Colle's fracture of his right hand. He underwent surgical control of lacerated liver injury. He required four units of whole blood transfusion in the perioperative period, and had an uneventful immediate postoperative recovery. On clinical examination, the patient was febrile, pallor and icterus were absent. Examination of the respiratory system revealed dull percussion note and diminished breath sound over the right chest, however there was no percussion tenderness.

A plain Chest X-ray (PA) view revealed a right-sided encysted pleural effusion [Figure 1]. Blood investigations showed normal hemoglobin but with leukocytosis (total leukocyte count- 14600/cu.mm), predominantly neutrophils. Liver function test was normal. Aspiration of the pleural fluid was carried out, macroscopically the fluid was turbid, mustard yellow in color, almost bile-like in appearance; hence, pleural fluid bilirubin level and other relevant studies were performed. Pleural fluid was exudative, cell count was increased (4300/cumm) with neutrophilic preponderance, adenosine deaminase was negative (10.7 U/l). On Gram staining showedthere was the presence of gram-negative bacilli but culture was negative, Ziehl-Neelsen stain was negative as well. Pleural fluid bilirubin was found to be 6.8 mg/dl. He was diagnosed as bilious pleural effusion with superadded bacterial infection. Ultrasound (USG) of the abdomen was done. It revealed two small hypo-echoic, round encysted collections, one in the left lobe of liver (segment VII) and the other in the subdiaphragmatic space, a fistulous tract was seen communicating between right pleural space and the collection in subdiaphragmatic space [Figure 2]. The findings were suggestive of biloma(s). However, percutaneous drainage

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DOI: 10.4103/0970-2113.159589

of the bilomas was not feasible as the collections were too small. Instead, the patient was managed conservatively i.e. pleural fluid was aspirated on two occasions 800 ml and 600 ml respectively and he was put on intravenous broad spectrum antibiotics (piperacillin-tazobactum and clindamycin). Follow-up CXR-PA view after 1 week showed minimal pleural effusion with residual pleural thickening. A repeat USG abdomen showed the two cysts had diminished significantly in size and the fistulous communication was no longer visualized. The patient underwent full recovery and was discharged well in a stable condition. On follow-up visit after 3 months, the patient was absolutely well; there were no pallor or icterus clinically. Examination of the respiratory system was also normal with the presence of normal breath sounds on both sides. Radiologically, the CXR was normal with minimal pleural thickening on the right side, USG of the abdomen also showed complete resolution of the biloma(s) and no fistulous tract could be identified [Figure 3].

DISCUSSION

Lungs and the liver are two adjacent viscera, hence lung–liver interface is not uncommon entity to pulmonologists. Clinically, possibility of a liver or subdiaphragmatic pathology has to be considered in all cases of right-sided pleural effusion. Liver abscess or subdiaphragmatic abscess are the common entities causing pleural effusion, but biloma with a fistulous communication with pleural space as an etiology of pleural effusion is extremely uncommon in the published literature.[1,3] Bilothorax can be iatrogenic or even rarely spontaneous. Laparoscopic or open surgical procedures of the hepatobiliary tree or blunt trauma are among the few causes of iatrogenic bilothorax.[3-8] On rare occasions, spontaneous bilio-pleural fistula can occur in patients with gall stones.[9] Rarely bile can reach directly to the bronchial tree by means of a broncho-biliary fistula and results in bilioptysis.[10] Bilothorax does not develop immediately following trauma, as the fistulous tract usually takes an average time of 2 weeks to mature and hence it is usually missed during initial emergency surgery.[11] In this case, the small biloma with the narrow fistulous communication between the subdiaphragmatic space and the pleura may not have been visualized during laparotomy and resulted in the leakage of bile causing bilious pleural effusion one month after the initial trauma. The proposed mechanism of formation of pleurobiliary fistula following injury to the biliary tree is formation of biloma and subsequent rupture of the biloma into the pleural cavity through the transdiaphragmatic route following the path of least resistance.[3,11] Passage of the bile through the connective tissue sheath of esophagus and great vessels to reach the posterior mediastinum and subsequent spillage from there into the pleural cavity or bronchus has been postulated as another theory for thoraco-biliary fistulas.[3]

Figure 1: Chest X-ray PA view showing right-sided encysted pleural effusion

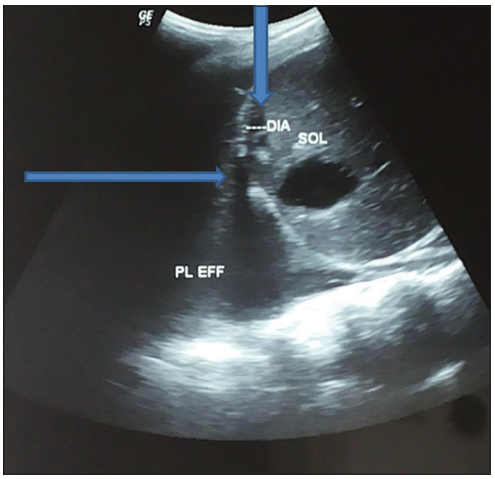

Figure 2: USG of abdomen showing two biloma, pleural effusion and the fistulous communication (Horizontal arrow) between the pleural space and the biloma in the subdiaphragmatic space (vertical arrow)

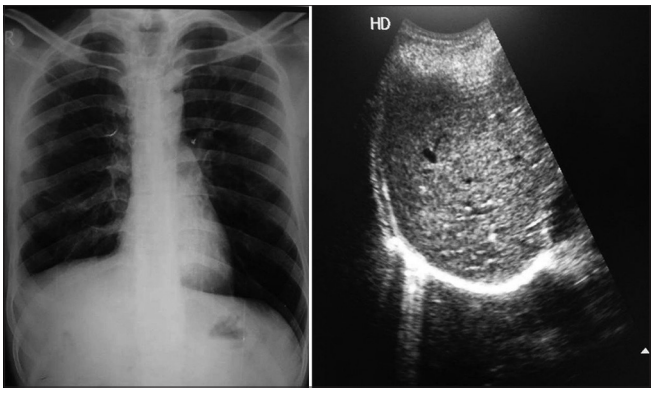

Figure 3: Follow-up Chest X-ray and USG of the abdomen after 3 months demonstrating complete radiological clearance of the pleural effusion and biloma(s), respectively

Diagnosis of bilothorax requires high index of clinical suspicion. On gross examination of the pleural fluid, greenish-yellowish tinge of the pleural fluid may give

hint toward the presence of bilirubin in the pleural fluid and the diagnosis is confirmed by estimation of bilirubin in the pleural fluid and a pleural fluid to serum bilirubin ratio greater than one.[12] Once the bilothorax is confirmed, the next step in diagnosis is to find out the fistulous communication between the pleural space and the biliary tree. Hepatic scintigraphy, endoscopic retrograde cholangiopancreatography (ERCP), or magnetic resonance cholangiopancreatography (MRCP) usually demonstrate the biliary leak and the fistulous communication.[13,14] USG of the abdomen is usually not a very sensitive tool for visualization of the fistulous tract but should always be done as a baseline test as it is inexpensive and at times can clinch the diagnosis by demonstrating the biloma and the fistulous tract and hence obviates the need for further invasive or expensive radiographic procedures as in our case.

Bilious pleural effusion poses therapeutic challenge to clinicians as bile is a very good nidus for bacterial infection.[3,6] In our patient, the pleural fluid was infected and it was manifested systemically as high grade fever. Increased pleural fluid cell count with neutrophilia and presence of gram-negative bacilli on Gram's stain also supported the evidence of the infection. The sequelae of bilo-thorax is fibrothorax as bile is a fibrogenic agent, hence delay in drainage of pleural fluid can rapidly give rise to a permanent state of compromised lung function.[4] Another rare but fatal complication of bilo-thorax is acute respiratory distress syndrome.[15] Early drainage of the pleural fluid either by intercostal tube or pig-tail catheter; or repeated therapeutic aspirations in case of multiple encysted fluids along with institution of broad spectrum antibiotics is the standard recommendation for management of bilo-thorax.[16,17] Transabdominal percutaneous drainage of the biloma has also proven to be beneficial in some cases.[3] Regarding the management of the bilio-pleural fistula, the fistulous tract usually heals spontaneously without any need for surgical intervention.[2,16-18]

In conclusion, though it is a rare entity, bilo-thorax is a diagnosis to should be kept in mind as a differential diagnosis in a case of right sided pleural effusion, especially after liver trauma or surgical intervention to the hepatobiliary tree. Early administration of broad-spectrum antibiotics with prompt therapeutic drainage of the pleural fluid will ensure a successful outcome.

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How to cite this article: Begum S, Mukherjee S, Biswas D, Misra AK, Ghosh P, Bhanja P. A rare pleural effusion in a young male. Lung India 2015;32:389-91.

Source of Support: Nil, **Conflict of Interest:** None declared.