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Case report

Transudative chylothorax from cirrhosis complicated by lung entrapment

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ABSTRACT

A patient with long-standing cirrhosis due to hepatitis C and hepatic hydrothorax was evaluated for increasing symptoms and presence of a large right pleural effusion. Thoracentesis revealed evidence of a chylothorax with rapid reaccumulation of pleural fluid. Repeat thoracentesis with manometry identified presence of entrapped lung which complicated treatment options. This is the first case report of a hepatic chylothorax with features of entrapped lung.

1. Introduction

A chylothorax is a well described pleural effusion characterized by the presence of triglycerides (>110 mg/dL) or chylomicrons in the pleural fluid. Malignancies, most commonly lymphoma, chronic lymphocytic leukemia, and metastatic cancer, are the leading causes of non-traumatic chylothorax [1]. Less frequently, chylothorax can be found in cirrhotics with chylous ascites where fluid leaks across the diaphragm into the pleural space; it may also be reported in conjunction with heart failure and the nephrotic syndrome [2,3]. Effusions due to chylous ascites are nearly always exudative though in a few cases they may be transudative by Light's criteria such as patients with amyloidosis, cirrhosis, nephrotic syndrome, superior vena cava obstruction, heart failure, and chylous ascites. Effusions related to chylous ascites are generally free-flowing and reaccumulate rapidly after a therapeutic thoracentesis. Lung entrapment results from an active pleural process, such as malignancy, infection, or inflammation that leaves the visceral pleura unable to fully expand after removal of an effusion. Lung entrapment is often suspected clinically but pleural manometry is required to establish definitive diagnosis [4–7]. We present a unique case of a non-inflammatory process, transudative chylothorax, complicated by lung entrapment.

2. Case report

A 67 year-old-female with chronic hepatic cirrhosis secondary to hepatitis C was evaluated in our pulmonary clinic for complaints of worsening exertional dyspnea and bilateral lower extremity edema. She had been diagnosed with a hepatic hydrothorax three years prior but had minimal ascites and concurrent pleural effusions were well managed with outpatient diuretic therapy consisting of furosemide and spironolactone. During the preceding month, her ascites had been recalcitrant to increases in diuretic therapy; she became progressively more dyspneic and had twice required therapeutic drainage of a recurring right sided pleural effusion.

On physical exam she was a frail, elderly female with diffuse spider angiomas on her upper torso and absent breath sounds over her right lower lung field. Abdominal distension was not noted, but she did have mild bilateral lower extremity edema. Repeat therapeutic thoracentesis was performed and was terminated when she experienced chest tightness and cough. Concurrent pleural manometry demonstrated a rapid drop in the pleural elastance at the time her symptoms began (Fig. 1).

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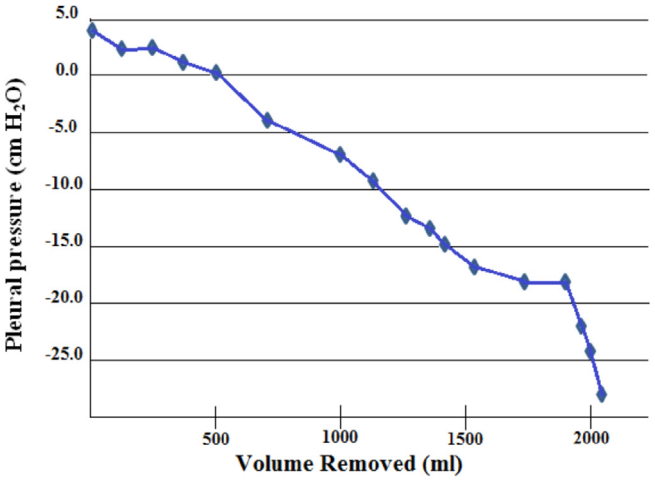

Fig. 1. Pleural manometry during thoracentesis. Legend: Pleural manometry demonstrating the presence of lung entrapment with relatively normal initial elastance curve followed by a sharp drop in pressure due to inability of lung to expand.

Subsequent post procedure ultrasound and chest X-ray showed no pneumothorax but did reveal a large residual effusion. The pleural fluid appeared turbid and milky while laboratory analysis was consistent with a transudative effusion with elevated triglycerides (Table 1).

There was no antecedent trauma, surgery, evidence of congestive heart failure, signs of nephrotic syndrome or any other obvious etiology for the chylothorax. Computed tomography scan of the chest revealed no evidence of malignancy, adenopathy or anatomical derangements. Abdominal ultrasound with Doppler flow showed no hepatic vein obstruction. In the ensuing weeks she underwent a steady clinical decline secondary to progressive liver failure. She developed hepatorenal syndrome and was unable to tolerate further diuretic therapy due to declining kidney function. She was ineligible for a transjugular intrahepatic portosystemic shunt due to her concurrent encephalopathy. After being evaluated and not deemed a candidate for orthotopic liver transplantation, she elected to have an indwelling pleural catheter placed and was discharged home with hospice.

3. Discussion

Classically described as a turbid, milky appearing fluid secondary to elevated chylomicrons and triglycerides, a chylothorax can also appear serous, serosanguinous or even bloody [3]. The heterogeneity in appearance of chylous effusions may lead to underdiagnosis as triglyceride levels are not routinely measured. Chylothoraces often occur secondary to disruption of the thoracic duct and have traditionally been classified by the underlying etiology: trauma; malignancy (most commonly lymphoma), idiopathic, and miscellaneous [8]. The vast majority of chylous effusions are exudative in nature.

Transudative chylothoraces have been associated with congestive heart failure, cirrhosis, amyloidosis, obstruction of the superior vena cava, and the nephrotic syndrome; however, the majority of these effusions have been associated with underlying cirrhosis [2,3,9]. The pathophysiology is believed to be elevated intra-abdominal pressures in conjunction with degenerative changes in splanchnic lymphatics resulting in lymph leakage into the peritoneal cavity. The transudative properties are likely secondary to dilution in concurrently forming portal hypertension ascites [10,11]. A preexisting anatomical defect allows this fluid to freely cross the diaphragm, a process expedited by the lower pressures found in the pleural cavity. Hence a chylous peritoneal effusion becomes a chylous hepatic hydrothorax [2]. In our patient, the rapid reaccumulation was further accelerated by decreased intrapleural pressures from her entrapped lung. Although there have been reports of transudative hepatic hydrothorax that subsequently became exudative following secondary bacterial infection, to the authors' knowledge there has been only one case report of such an event involving a hepatic chylothorax [12]. The reason for the low propensity for secondary infection is presumed to be the inherent bacteriostatic nature of chyle [13].

The transudative effusion in the setting of lung entrapment is a unique feature in this case. Patients with an entrapped lung are usually symptomatic on presentation and have exudative effusions in the vast majority of cases [5]. Chest discomfort often occurs during thoracentesis despite a considerable amount of residual fluid [14,16]. There have been no prior reported cases of lung entrapment following from a transudative chylothorax. Trapped lung results from previous pleural inflammation resulting in fibrosis and scarring of the visceral pleura. The inciting event is typically an infectious process such as pneumonia although alternate etiologies such as hemothorax, pneumothorax, thoracic cavity interventions, uremia and rheumatoid pleuritis have been reported [5–7]. The inability of the affected lung to expand results in excessive negative intrathoracic pressures with subsequent development of an effusion ex vacuo [15]. These effusions are always transudative and patients are typically asymptomatic or may complain of dyspnea due to the restrictive effects of the unexpandable lung [5].

The unexpandable lung is diagnosed via pleural manometry. This involves intermittent measurement of pressure in the pleural space during thoracentesis. The data points are mapped as a change in pressure over change in volume to create a pleural elastance curve. While several different methods of measuring the

Table 1
Comparison of pleural fluid studies from 2010 to 2013.

Year	2010	2013	Interpretation (per lights criteria [8])
Color	Straw	Cream, turbid	
pH	7.71	7.69	
Pleural fluid (PF) protein (g/dL)	1.5	0.5	
Serum total protein (g/dL)	7.1	6.7	
PF/Serum protein ratio	0.21	0.07	Ratio > 0.5 classified as exudative
PF Lactate Dehydrogenase (LDH) (IU/L)	87	30	LDH > 2/3s the upper limit of normal (148.5 IU/L)
Serum LDH (IU/L)	205	151	
PF/Serum LDH ratio	0.42	0.19	Ratio > 0.6 classified as exudative
PF Glucose (mg/dL)	90	131	
PF Triglyceride (mg/dL)	5	599	Triglyceride > 110 mm g/dL classified as a chylothorax
PF Cholesterol (mg/dL)	Not measured	34	
Bacterial Culture	Negative	Negative	

intrapleural pressure, the concept of measuring the change in pulmonary elastance is the same for all methods [5,14,16] Three classic elastance curves were described by Light et al. [17]. In a normal pressure curve there is an initial positive pressure with an almost linear drop in pressure as volume is removed. Lung entrapment produces an initially normal curve with a sharp negative descent once the lung is unable to expand further. Patients with trapped lung demonstrate an initially negative pleural pressure with a steady, rapid drop in pressure during thoracentesis. It has been proposed that the longstanding active inflammation that results in lung entrapment leads to development of pleural fibrosis and subsequent trapped lung that persists after resolution of the initial insult [5,15]. This would imply that lung entrapment and trapped lung are on a spectrum of pleural disease rather than distinct entities as initially suggested by Light's pleural manometry data.

4. Conclusion

There are several unique characteristics of this case that have not been previously reported. Transudative chylothoraces are exceedingly rare entities most commonly occurring in patients with chronic cirrhosis and hepatic hydrothorax. They have never been previously described in association with entrapped lung. Though an extensive workup was performed, no inflammatory etiology was ever discovered. The presence of the entrapped lung appearing at the same time as the identification of the chylothorax suggests it may have contributed to the underlying process. Alternatively, it is possible the patient had another inflammatory pleural process that developed between her initial thoracentesis and subsequent follow up that predisposed her to development of an entrapped lung. Clinicians should have a low threshold to measure triglyceride levels in patients with known hepatic hydrothorax and consider the use of manometry during thoracentesis.

Conflict of interests

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"The view(s) expressed herein are those of the author(s) and do not reflect the official policy or position of Brooke Army Medical Center, the U.S. Army Medical Department, the U.S. Army Office of the Surgeon General, the Department of the Army and Department of Defense or the U.S. Government."

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