CASE REPORT

Transudative chylothorax: an uncommon illness due to a common cause

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SUMMARY

Chylothorax is defined as presence of chyle in the pleural space. It is commonly associated with cardiothoracic surgery, trauma, malignancy or some benign disorders. Transudative chylothorax is uncommon. A 52-year-old man presented with bilateral chylothorax with preceding history of blunt trauma to the chest. On further evaluation, he was diagnosed to have a transudative chylothorax due to cirrhosis of liver with coexisting chylous ascites and evidence of peritoneopleural communication. The patient was managed with diuretics followed by chemical pleurodesis with iodopovidone.

BACKGROUND

Chylothorax is defined as presence of chyle in the pleural cavity. It is classically described as an exudative milky white pleural effusion. The causes of chylothorax include trauma (surgical or non-surgical), lymphomatous or non-lymphomatous malignancies, and several benign disorders such as infections (tuberculosis, filariasis), sarcoidosis and others.¹ Chylothorax-related effusion is generally exudative; a transudative chylothorax is uncommonly encountered. Herein, we describe a patient with a chylothorax due to an uncommon cause.

CASE PRESENTATION

A 52-year-old man presented with dry cough, pleuritic chest pain and progressive shortness of breath of 2 months' duration. The patient first noticed swelling in both lower limbs about 3 months back. After a month, he developed dry cough and breathlessness that gradually increased in intensity. He also complained of pain in the right side of the chest that got aggravated with inspiration. He preferred to sleep on his right as he had less breathlessness and chest pain in that position. There was no fever, loss of appetite or weight. The patient did not smoke but consumed 80–100 g of alcohol daily for the preceding 10 years. He had hypertension and hypothyroidism, which were treated with amlodipine and levothyroxine, respectively. Three months prior to his current illness, he had suffered blunt trauma to the chest and abdomen in a road traffic accident. He had complained of chest pain at that time which was managed with 5 days of analgesics; no imaging of the chest and abdomen was performed.

On examination, the patient was alert. The pulse rate was 90 per minute, respiratory rate 18 breaths per minute, blood pressure 110/70 mm Hg

and oxygen saturation was 98% by pulse oximetry on breathing ambient air. There was no pallor, icterus, cyanosis or clubbing. Jugular venous pressure was not raised. There was bilateral pitting pedal oedema. There was no peripheral lymph node enlargement. Fundus examination did not reveal any abnormality. A dull percussion note was present in the left infrascapular area accompanied with decreased intensity of breath sounds. There was presence of an intercostal drainage tube on the right side (inserted in another hospital the patient had visited) that drained milky white pleural fluid. The abdomen was mildly distended with free fluid (shifting dullness present). Liver and spleen were not palpable and there were no abnormally distended veins over the abdomen.

INVESTIGATIONS

Laboratory investigations revealed normal blood counts, plasma glucose, renal function tests and urine examination (table 1). Liver function tests revealed mildly raised liver enzymes (<3 times the upper limit of normal) and a reduced albumin to globulin ratio. Hepatitis B surface antigen and IgG against hepatitis C virus were absent in serum and ELISA for HIV was negative. Thyroid-stimulating hormone level was high (9.21 µIU/mL) with normal serum T3 and T4 levels. Arterial blood gases revealed respiratory alkalosis with metabolic compensation.

Chest radiograph showed bilateral pleural effusion with an intercostal tube in the right pleural cavity. Pleural fluid from left pleural cavity and the ascitic fluid were aspirated for investigations. Pleural fluid from the left side and the ascitic fluid had serous appearance. Pleural fluid analysis from both sides revealed a transudate effusion with elevated triglyceride concentration (table 2). His ascitic fluid evaluation showed low serum ascitic fluid albumin gradient with raised triglyceride. A diagnosis of bilateral transudative chylothoraces with chyloperitoneum was made.

DIFFERENTIAL DIAGNOSIS

- ▶ Cirrhosis of liver
- ▶ Congestive heart failure
- ▶ Chronic constrictive pericarditis
- ▶ Superior vena cava obstruction
- ▶ Traumatic chylothorax
- ▶ Lymphoma
- ▶ Tuberculosis
- ▶ Filariasis

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Table 1 Haematological and biochemical parameters of the index patient

Parameter	Value
Haemoglobin (g/dL)	14
Total leucocyte count ($\times 10^9$ cells/L)	8.5
Platelet count ($\times 10^9$ cells/L)	335
Urea (mg/dL)	24
Creatinine (mg/dL)	1.0
Sodium (mEq/L)	139
Potassium (mEq/L)	4.0
Calcium (mg/dL)	9.2
Phosphate (mg/dL)	2.0
Bilirubin (mg/dL)	0.5
Aspartate transaminase (U/L)	87
Alanine transaminase (U/L)	62
Alkaline phosphatase (U/L)	149
Protein (g/dL)	6.7
Albumin (g/dL)	2.8

Adenosine deaminase levels in the pleural fluid from the right and left sides were 21 U/L and 29 U/L, respectively, indicating a low probability of tuberculosis. Magnetic resonance lymphangiography did not show any leak from the thoracic duct thereby excluding traumatic aetiology of chylothorax. Echocardiography was normal. Microfilaria antigen was not detected in serum. Ultrasonography of the abdomen showed an enlarged liver with coarsened echotexture. Contrast-enhanced CT (CECT) scan of the chest and abdomen revealed presence of bilateral pleural effusions with no lymph node enlargement. The liver was enlarged, heterogeneous in attenuation with nodular outline; portal vein diameter was 14 mm. Few collateral vessels were seen in perigastric, perisplenic and mesenteric areas. Fibroscan of liver revealed a liver stiffness measurement of 16.8 kPa with an IQR, 2.8 kPa, success rate 100% consistent with cirrhosis of liver. Upper gastrointestinal endoscopy showed grade II oesophageal varices. Tc99m sulphur colloid peritoneopleural study revealed peritoneopleural communication bilaterally, with a higher transmigration of the tracer on the left side (figure 1). A diagnosis of cirrhosis of liver (Child-Turcotte-Pugh score B) with chylous ascites and bilateral chylous pleural effusions was made.

TREATMENT

There was a daily drainage of 200–300 mL of milky white fluid from the right intercostal tube. The patient was administered propranolol, furosemide, spironolactone, subcutaneous unfractionated heparin and pantoprazole. The dose of levothyroxine was increased. Dietary fats were restricted and dietary

supplementation with medium-chain fatty acids was given. Chest drain output reduced in 2 days. Pleurodesis was performed successfully with iodopovidone followed by removal of the intercostal drainage tube. The effusion in the left hemithorax and the peritoneal cavity did not require drainage.

OUTCOME AND FOLLOW-UP

The patient reported to be doing well during the follow-up visit at 1 year with no recurrence of ascites. Chest radiograph showed minimal bilateral pleural effusion and right-sided pleural thickening.

DISCUSSION

The index case highlights that chylothorax may be transudative, have a serous appearance and can occur due to cirrhosis of liver. Pleurodesis may be performed successfully in such a case with iodopovidone.

Chylothorax occurs due to obstruction or disruption of the thoracic duct, leakage of chyle from lymphatic masses or lymphatic malformations or by the transdiaphragmatic migration of chylous ascitic fluid. The diagnosis of chylothorax requires the demonstration of chylomicrons in pleural fluid. As this requires a lipoprotein electrophoresis, which is cumbersome, pleural fluid triglyceride level >110 mg/dL (present in 85 % of patients) is generally used to diagnose chylothorax.² Chylothorax is unilateral in most patients, but may be bilateral in about 20% of the cases.³ Chylothorax has been traditionally considered as a milky white lymphocytic exudative effusion.^{4,5} However, in a retrospective study of 74 chylothorax cases, only half of the cases had a milky appearance, and 14% cases were transudative.⁵ The index case had bilateral chylous pleural effusions that were transudative; the fluid aspirated from the left pleural cavity had a serous appearance.

Transudative chylothorax has been described scarcely. In a systematic review in 2005, 20 cases of transudative chylothoraces were included.⁶ Cirrhosis of liver was the most common cause accounting for about 40% of the cases. Other causes of transudative chylothorax included heart failure, nephrotic syndrome, superior vena cava thrombosis and others. In one study, all cirrhotic chylothoraces were transudates according to Light's criteria.⁷ Most patients with chylothorax in the setting of cirrhosis also have a coexisting chylous ascites. Chylous ascites is a well-known complication of cirrhosis of liver. In a systematic review of 194 patients with atraumatic chylous ascites, 11% were due to underlying cirrhosis.⁸ However, chylothorax has been less frequently reported in this setting. It has been postulated that portal hypertension with resulting disruption of the serosal lymphatics causes chylous ascites in cirrhosis. Chylothorax results from subsequent transdiaphragmatic spread of chylous ascites. As a result of dilution with the pre-existing effusions (ascites and pleural effusion) due to portal hypertension, these chylous effusions have transudative characteristics.⁷ An alternative explanation for the formation of chylothorax is increased formation of lymph with increased pressure and flow rate in the thoracic duct in the presence of portal hypertension and chylous ascites, as observed in animal models of cirrhosis.⁹ In the study by Romero *et al*, peritoneopleural shunting was observed on scintigraphy in patients with cirrhosis-associated chylothorax.⁷ The index patient had cirrhosis with coexistence of chylous ascites and bilateral chylothoraces. A communication between the peritoneal and bilateral pleural cavities could be demonstrated pointing towards transmigration of fluid as the underlying mechanism. Further, we excluded any obstruction

Table 2 Pleural fluid characteristics of the index patient

Characteristic	Right-sided pleural fluid	Left-sided pleural fluid	Serum
Appearance	Milky white	Serous	
Total protein	3.4 g/dL	3.1 g/dL	6.9 g/dL
Sugar	103 mg/dL	112 mg/dL	150 mg/dL
Triglyceride	366 mg/dL	331 mg/dL	136 mg/dL
Cholesterol	57 mg/dL	61 mg/dL	173 mg/dL
Adenosine deaminase	21 IU/L	29 IU/L	
Cytological examination	No malignant cell seen

Figure 1 Tc99m sulphur colloid peritoneopleural study showing (A) tracer in the abdomen at the beginning of the study (black arrows point towards the markers at the sternal notch and xiphisternum), (B) anterior view of the thorax showing the presence of the tracer in the left hemithorax at 1 hour, (C) anterior view at 24 hours showing the presence of tracer bilaterally, more in the left hemithorax and (D) posterior view of the thorax at 24 hours.

or disruption of the thoracic duct using CECT of the chest and abdomen and MR lymphangiography.

The management of chylothorax involves treatment of the underlying cause, reduction of lymph flow by dietary approaches (high-protein low-fat diet, fat-free diet, diet with medium-chain fatty acids, complete fasting with total parenteral nutrition) and somatostatin analogues; pleural drainage; pleurodesis; and, thoracic duct embolisation/disruption or surgical ligation.^{3 10 11} In chylothorax due to cirrhosis, conservative management is usually advocated and consists of repeated thoracentesis, diuretics, dietary interventions and octreotide. Dietary intervention in the form of a fat-free or low-fat diet reduces lymph formation.¹² In case of no response, total parenteral nutrition may be employed. Medium-chain fatty acids should be supplemented as they are absorbed directly from the intestinal epithelial cells into the bloodstream without traversing through the lymphatic system. Somatostatin analogues such as octreotide reduce the overall intestinal secretion and absorption. Tube thoracostomy and pleurodesis have also been described in the treatment of cirrhotic chylothorax, although they should be used only if conservative measures fail.¹³ In the index case, we employed a low-fat dietary regimen with medium-chain fatty acid supplementation along with diuretics and propranolol, which successfully reduced the intercostal drain output. Had the intercostal drain not been placed prior to presentation to us, we would have avoided the same. However, as the tube was already in place, we attempted pleurodesis with iodopovidone, which proved to be successful.^{14 15} Transjugular intrahepatic portosystemic

shunting (TIPSS) is another effective treatment option for refractory chylothorax in patients with cirrhosis. In a recent systematic review, TIPSS has been described as a successful treatment in 10 cases of cirrhotic chylothorax refractory to conservative management.¹⁶

Learning points

- ▶ A chylothorax may be serous in appearance and may show features of a transudate.
- ▶ A transudative chylothorax may be encountered along with coexisting chylous ascites in patients with cirrhosis of the liver.
- ▶ Chylothorax in cirrhosis occurs due to transmigration of chylous ascitic fluid across the diaphragm.
- ▶ Diuretics and fat-free diet are effective in reducing the lymph flow in patients with chylothorax due to cirrhosis.
- ▶ Although not the preferred option, intercostal tube drainage with chemical pleurodesis with povidone-iodine is effective in the management of a cirrhotic chylothorax.

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