Idiopathic Massive Bilateral Chylothorax: A Case Report

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Abstract

Introduction: Chylothorax is an uncommon medical condition caused by the accumulation of chylous fluid in the pleural space. Chylothorax has no predilection for sex or age. The prevalence after various cardiothoracic surgeries is 0.2% to 1%. Mortality and morbidity rates are around 10%. Respiratory distress may occur due to compression of the lung by the accumulated fluid. Management and approaches to treating the condition require multidisciplinary therapy, starting from non-pharmacological, pharmacological, to interventional management.

Case Report: A 57-year-old male patient was referred to the emergency room with chief complaint of shortness of breath. Reduced breathing sound on both lung fields. No previous history of cancer or thoracic surgery were found. X-ray examination had found bilateral pleural effusion. Thoracocentesis and pleural fluid analysis was performed with a total of 6800 cc serosanguinous, whitish fluid was extracted from both of the lung. The patient was diagnosed with chylothorax. Lymphangiography and embolization was performed on the leak on left thoracic duct (T10) from right lymph node. Antibiotic was also given to treat the community acquired pneumonia that could be one of the possible etiology on this patient. Dietary modification with low fat diet and Ocreotide was also given to this patient as one of the treatment modalities.

Discussion: The diagnosis of Chylothorax on this patient was established based on pleural fluid analysis and evidenced by lymphangiography examination by the presence of a leak in the thoracic lymphatic duct. Various modalities to diagnose this condition have been carried out with inconclusive results. Non-pharmacological, pharmacological and radiological interventions with embolization through lymphangiography are proven to be able to stop leaks and reduce symptoms in this patient.

Conclusion: Chyle leak to the pleural space may compress the lung and cause respiratory distress. Combination of thoracocentesis, embolization of the leakage, dietary intake modification and administration of ocreotide may help prevent further chylous fluid accumulation.

Keywords: chylothorax, embolization, lymphangiography, thoracocentesis.

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Introduction

Chylothorax is an uncommon medical condition characterized by accumulation of chyle in the pleural space. Numerous etiologies for the case had been suggested, with the most common being previous history of thoracic surgery. Chylothorax may range from relatively asymptomatic into potentially life-threatening condition due to the resulting chyle build up that compresses the lung and leading to respiratory arrest.(1) In this case, we present a case a massive bilateral chylothorax with respiratory distress.
Case Report

Mr. R, a 57-year-old patient presenting to the emergency room with chief complaint of shortness of breath. The patient had felt shortness of breath approximately 3 months ago. The shortness of breath had worsened about a week ago. The patient had also complained chest pain during breathing in. Previously, the patient had also reported occasional, shortness of breath for about 30 years ago. During the time period, the patient had never sought medical care. The patient had reported that his shortness of breath worsened during physical exertion. The patient denied any history of swelling extremities or shortness of breath when lying down on his back. The patient occasionally had dry coughs. No previous history of hypertension, diabetes mellitus, cardiovascular disease, coronavirus-19 infection, and asthma. The patient does not smoke nor consume alcohol.

The referring hospital had decided to perform thorax x-ray scan and had found bilateral pleural effusion. Afterwards, thoracocentesis was performed on the patient; the pleural fluid was serous, whitish; hence, the referral of the patient to our institution. During the physical examination, the patient was fully alert and moderately ill. Hypotension (95 / 70 mmHg) and increased breathing rate (26 times/ min) were found on the patient. The rest of the vital signs were within normal limits. Peripheral oxygen saturation was decreased on the patient (94 – 95%, supplemented with 3 lpm nasal cannula). Body mass index (BMI) were within normal limits; the patient’s height and weight were 155 cm and 47 kg respectively. The patient was within normal weight (body mass index of 19.6 kg/m²).

On auscultation, breathing sounds on both sides of the lung were reduced; wheezing was found during auscultation. No rhonchi were found on any part of the chest during auscultation.

Complete blood count during the admission were within normal limits. Differential count had noted neutrophilia (83.4%) during the admission. Analysis of the pleural fluid had noted high amount of protein (4.32 g/dl) and glucose (108 mg/dl) an also Triglyceride (247 mg/dl). On blood gas analysis, compensated metabolic alkalosis with respiratory acidosis was found on the patient (treated with FiO2 37% with supplementary oxygen delivered through nasal cannula with flow rate of 4 lpm). Bilateral pleural effusion was found on thorax x-ray during admission (Figure 1). The microbiology assessment shows there is no AFB (Acid Fast Bacteria) indicating MTb (Mycobacterium tuberculosis) infection was found. Sputum and pleural fluid culture and cytology was also negative.
Total 6800 cc of pleural fluid were extracted from both of the lung (Figure 2). On the 5th day of admission thorax lymphangiography and embolization were performed by intervention radiology department. Lymphangiography had found leakage to the horizontal direction on left thoracic duct (T10) (Figure 3). Embolization was performed with glue : lipiodol (1 : 6) 1.75 cc from right inguinal lymph node and flushed afterwards with D5% 6 cc. The leakage had been reduced (confirmed with fluoroscopy and DSA examination). Additional CT scan had found pneumonia with inferior lobe bronchiectasis with bilateral fluid accumulation (Figure 4). Based on the patient factors, the Pneumonia Severity Score (PSI) was 67 (risk class II). The sputum culture results were negative.

The patient was diagnosed with bilateral chylothorax and community-acquired pneumonia. Empirical antimicrobials were given to the patient to treat the community acquired pneumonia that possibly could be one of the etiology of his condition. Low fat diet and Octreotide (3 x 100 mcg) were prescribed to prevent further chyle buildup on pleural space. We still follow the patient up until now and he is remain stable with no history of worsening symptomps.

Figure 1. Bilateral pleural effusion found on thorax X-ray during admission

Figure 2. Extracted pleural fluid from both right and left hemithorax
Discussion

Chylothorax is defined as a collection of chyle on thoracic cavity. Etiology of the chyle leak from thoracic duct were broadly classified into traumatic and non-traumatic (Figure 5). The contents of chyle may result in differing clinical signs. A chylothorax has been defined as fluid with either or both triglycerides >110mg/dL and the presence of chylomicrons, which are considered the gold standard for diagnosis. Rapid loss of chyle may cause symptoms such as hypovolemia and respiratory issues due to the pleural space being filled with fluid. In sufficiently severe cases, some patient may present with signs of malnutrition due to the loss of proteins, fats, and vitamins. Electrolyte loss and the resulting electrolyte abnormalities detected during the examination may be found. Chylothorax may occur due congenital, traumatic, neoplastic, or other causes; the bulk of the cases occur as a complication from previous thoracic surgery. In this case, it is assumed that the patient had no known cause of chylothorax. No previous reports of malignancy or thoracic surgery prior to the admission. The idiopathic chyle leak of the patient was masked due to the frequently prescribed Symbicort nebulization during the shortness of breath. Hence, the patient’s presentation with noticeable respiratory distress that remained unresolved after nebulization. Notably, the patient had no previous history of asthma, chronic obstructive pulmonary disease (COPD), or other medical conditions that require the use of inhaled corticosteroids during shortness of breath. No previous studies were found in regards to the effects of prior inhaled corticosteroid use with the risk of chylothorax.
Chylous fluid leaking into pleural space treated based on the nature of the leak and the presenting symptoms. In cases with the risk of respiratory distress, thoracocentesis to drain the fluid and analyze the content of the fluid often function as first-line therapeutic and diagnostic effort, respectively. Thoracic duct embolization may be performed after identification of chyle leakage (Figure 6). Outcomes are generally favorable post-embolization.\(^{(3)}\) The chyle leak on T10 was discovered during the lymphography on the 5th day of treatment. Afterwards, the patient was scheduled to receive embolization of thoracic duct on T10 in order to occlude the leakage and prevent further chyle leak to the pleural space. The diagnosis of chylothorax was confirmed from the gross look of milky appearance (Figure 2) and with the pleural fluid analysis is from the high protein, glucose content of the fluid, and also high number of Triglyceride on the pleural fluid.

Another cause of milky appearing pleural effusion is a pseudochylothorax, also known as a cholesterol effusion and chyliform effusion, which is a cholesterol-rich fluid associated with chronic inflammatory disorders. The clinical definition of a pseudochylothorax is the combination of a milky pleural effusion, pleural cholesterol level greater than 200 mg/dL, pleural triglyceride level typically below 110 mg/dL, a pleural cholesterol/triglyceride ratio of greater than one, and often the presence of cholesterol crystals seen on microscopy.\(^{(2)}\) The thoracic duct functions to transport chyle back to the blood-stream. In a normal adult, the thoracic duct transports between 1.5 and 4 l of chyle per day. Chyle is produced in the small intestine via the capillary filtrate, which consists of fluids, electrolytes, peptides and lipoproteins, and intestinal absorption and secretion by enterocytes that line the intestinal epithelium. Long-chain triglycerides in the diet are converted into chylomicrons and very-low-density lipoproteins. Chyle is mainly composed of chylomicrons (a form of triglycerides that

Figure 4. Thorax CT scan found pneumonia with inferior lobe bronchiectasis with bilateral fluid accumulation after Thoracocentesis was performed.
are large spherical proteins that transport large amounts of dietary fat absorbed by the small intestine to the lymph from the enterocytes), lymphocytes (predominately T-lymphocytes), electrolytes, immunoglobulins, albumin, fibrinogen, glucose and fat-soluble vitamins.\(^{(4)}\) Thoracocentesis in this case is only a temporary intervention in order to improve the pulmonary function of the patient. Due to the massive amount of chyle leakage (more than 5,000 mL in total in less than a week), the patient was opted to receive lymphangiography to detect the location of the leak and perform embolization afterwards to reduce the amount of leakage \(^{(5)}\).

Somatostatin and its analogue exert a broad spectrum of inhibitory actions in many organs, including the central nervous system, the pituitary gland, the liver and the pancreas, as well as the gastrointestinal tract.\(^{(6)}\) The inhibition of serotonin and other intestinal peptides produces an increase in water absorption and intestinal transit and a decrease in pancreatic-duodenal secretion. More importantly, the resistance to splenic blood flow increases, and intestinal arteriolar flow decreases, in turn reducing lymphatic flow.\(^{(3)}\)

Surgical therapy typically requires identification of the leak, which can be accomplished by lymphangiography or lymphoscintigraphy. Once the site of extravasation is visualized, a minimally invasive technique such as percutaneous embolization.\(^{(4)}\) In cases where conservative management failed, surgical intervention in the form of video-assisted thoracoscopic (VATS) thoracic duct ligation was advised.\(^{(7)}\) The possible etiologies of chylothorax are on Figure 5.

**Conclusion**

Idiopathic chylothorax is a rare disorder. Massive fluid buildup on the pleural space compromises the respiratory function of a patient. Thoracocentesis to remove the excess fluid was used as a temporary solution until embolization and occlusion of the thoracic duct leak were addressed. Massive chyle leak may lead to weight loss and respiratory distress due to the loss of chyle to the pleural space and its subsequent compression of the lung, affecting its ability to expand. Management

![Figure 5. Etiology of Chylothorax](image-url)

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of a chylothorax requires a multidisciplinary approach, employing medical therapy and possibly surgical intervention.

![Figure 6. Proposed algorithm for stepwise conservative and surgical management.](image)

Consent

Written informed consent was obtained from the patient for publication of this case report.

References