

A Case Series of Spontaneous Chylothorax in Lymphoma

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ABSTRACT

Introduction: Chylothorax is a rare condition originating from lymphatic system damage as an accumulation of chyle in the pleural space. It can be classified into spontaneous or non-traumatic, traumatic, and idiopathic. A spontaneous chylothorax is primarily caused by neoplasm, such as lymphoma.

Case Presentation: Herein, we report two cases of chylothorax presenting as massive pleural effusion in two patients of a 23-year-old male with nodular sclerosis type Hodgkin's lymphoma and a 31-year-old female with metastatic lymphadenopathy of undifferentiated carcinoma from non-Hodgkin's lymphoma.

Conclusion: A spontaneous chylothorax is a rare condition that may lead to a life-threatening event when it is not properly and promptly treated.

INTRODUCTION

Chylothorax is a condition where there is a chyle deposition in the pleural cavity. This rare condition may affect both males and females regardless of age [1]. The word chyle itself is derived from the Greek word "Chylous", which means juice. Leakage of chyle into the pleural cavity has resulted from damage to the thoracic duct. Chyle is a milky fluid produced in the lacteal system of the intestine [1,2].

The cause of chylothorax is categorized into non-traumatic or spontaneous, traumatic, and idiopathic. Non-traumatic or spontaneous chylothorax is mostly caused by neoplastic chylothorax. Several types of cancer such as lymphoma, chronic lymphoid leukemia, lung cancer, or metastatic carcinoma may have the clinical presentation of chylothorax. However, there is a considerable decline in chylothorax incidence in patients with lymphoma. It is perhaps due to early diagnosis and treatment of underlying disease, preventing the late complication of chylothorax [3].

Since 2013, we have encountered 13 cases of chylothorax in our tertiary center, dr. Moewardi Hospital, Surakarta. In this report, we describe the management of chylothorax by emphasizing radiologic findings of two patients who experienced different outcomes.

CASE PRESENTATION

We retrospectively report two of the 13 chylothorax cases in our tertiary center, dr. Moewardi Hospital, Surakarta during the period of 2013 to 2019. These two cases were the patients who have complete workup including imaging modality. The first case is a 23-year-old male with nodular sclerotic type Hodgkin's lymphoma. He presented with shortness of breath, chest discomfort, prolonged cough with milky sputum, and no smoking history. Biopsy of the lesion obtained nodular sclerotic type Hodgkin's lymphoma. Contrast-enhanced Chest Computed Tomography (CT) Scan demonstrated superior mediastinal mass and bilateral milky effusion with collapsed left lung (**Figure 1**). Milky effusion was drained with the amount of 6,250 ml from the left and 2,100 ml from the right hemithorax (in 8 days), with 845–1457 mg/dl level of triglyceride. Bilateral productive massive milky effusion containing a high level of triglyceride led to the diagnosis of chylothorax. The patient, however, refused chemotherapy and his dyspnea worsened. He ultimately died 1 year following conservative treatment.

The second case is a 31-year-old female with metastatic lymphadenopathy from undifferentiated carcinoma (non-Hodgkin's lymphoma). She was admitted with breathlessness, chest pain, prolonged cough with milky sputum, and no smoking history. Chest X-Ray and

Figure 1. Imaging of a 23 y.o male with nodular sclerotic type Hodgkin’s lymphoma. Axial (A) and coronal (B) sections of contrast-enhanced chest CT-Scan of shows a superior mediastinal mass (black arrow) with bilateral effusion pleura (blue arrow). A chest drain is inserted on the left chest wall into the left pleural cavity.

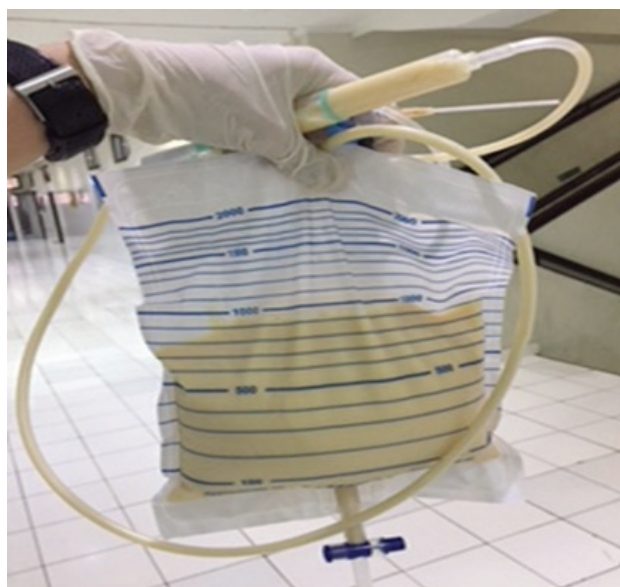
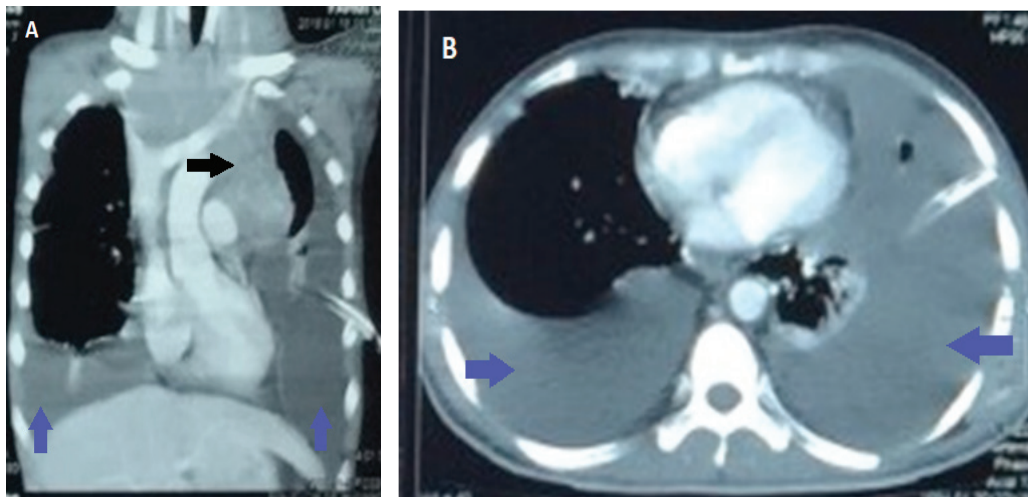


Figure 2. Drainage of pleural effusion shows milky chylous liquid.

contrast-enhanced CT scan delineated right upper mediastinal mass with left pleural effusion. The biopsy demonstrated non-Hodgkin’s lymphoma and bilateral supraclavicular lymph nodes revealed metastatic undifferentiated carcinoma. Continuous chest drainage showed milky effusion with a total of 1,900 ml containing 357 mg/dl level of triglyceride (**Figure 2**). The patient subsequently underwent chemotherapy and was discharged in a stable condition.

DISCUSSION

Hodgkin’s lymphoma is an uncommon B-cell malignant neoplasm [4]. It represents approximately 11% of all lymphomas seen in the United States and is classified into nodular sclerosis, mixed cellularity,

lymphocyte depletion, and lymphocyte-rich [5]. During the initial presentation in 21% of HL cases, the most common type is nodular sclerosis [6], as presented in our first case. Nodular sclerosis Hodgkin’s lymphoma with spontaneous chylothorax complication is rare [4,7].

The evaluation of chylothorax depends on the suspected cause and the availability of resources. Radiographic finding from both patients in our center was unspecific for chylothorax. Chest X-ray finding of chylothorax showed homogenous opacity filling costophrenic and cardiophrenic angles [8]. CT scan is more sensitive than chest X-ray for diagnosing chylothorax. Due to the high amount of fat content, it is seen as a low attenuation tubular area in the posterior mediastinum. CT scan may also show the possible etiology of chylothoraces such as mass or obstructive lesion in the posterior mediastinum, malignancy in the thorax cavity, or evidence of trauma.

The clinical features of chylothorax depend on the etiology. Small chylothorax can appear without symptoms and be detected incidentally. Large chylothorax normally presents with signs and symptoms resulting from the mechanical effect of compression on lung expansion, as present in both of our cases. Decreased breath sounds and dullness to percussion may be present on the physical examination depending on the size and the site of fluid [8].

In general, chylothorax in the setting of lymphoma can be treated with conservative management, radiation, pleurodesis, pleuroperitoneal shunt, or surgery. Even though conservative management including dietary modifications, such as a low-fat diet or total parenteral nutrition, can lower the production of chyle, chylothorax will reappear if the primary cause is left untreated [9]. Thoracentesis can be beneficial for the initial management of malignant chylothorax as it can immediately relieve respiratory symptoms [10]. Meanwhile, surgical ligation of the thoracic duct as a treatment option for traumatic

chylothorax in the setting of malignancy is not commonly applied because such patients mostly are medically inoperable [11]. Chemotherapy could be the first choice of treatment for malignancy chylothorax, as shown in our cases [10–12]. Different outcomes may present in both of our patients. Our second case undergoing chemotherapy was stable during treatment while the first case died without chemotherapeutic administration.

CONCLUSIONS

A spontaneous chylothorax is a rare condition that may lead to a life-threatening event when it is not properly and promptly treated. Radiologic imaging plays an important role as a diagnostic modality. CXR could be initially performed to identify a pleural effusion leading to a chest drainage procedure despite its little diagnostic value. Chest CT scan may be effective to identify the site and the etiologies of chylous leakage such as mediastinal lymphoma as well as to evaluate post WSD procedure. Accurate identification of the etiology of chylothorax is an indicator in determining the optimal course of treatment for individuals. Most spontaneous chylothorax cases can be successfully handled by addressing the underlying condition in conjunction with conservative treatment. They often require surgical therapy to achieve resolution.

DECLARATIONS

Competing of Interest

The authors declare no competing interest in this study.

Acknowledgment

Not applicable

Ethics approval and consent to participate

This report has already been approved by the Health Research Ethics Committee of Moewardi General Hospital 1.197/X/HREC/2020 issued on October 21, 2020.

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