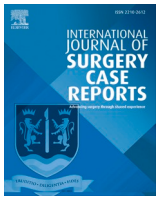
International Journal of Surgery Case Reports 102 (2023) 107827

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr



Case report

Indonesian female with bilateral chylothorax and mediastinal non-Hodgkin lymphoma: A case report

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ARTICLE INFO

Keywords:
Bilateral chylothorax
cancer
Mediastinal
Non-Hodgkin lymphoma

ABSTRACT

Background: Bilateral chylothorax is leakage and accumulation of lymph fluid in the pleural space on both sides of the lung and in non-traumatic cases, caused mainly by lymphoma.
Case presentation: An Indonesian female, 34 years old, complained of short breath, cough, and swelling in several areas (neck, breast, and hands). Chest X-ray and thorax CT scan showed the anterior mediastinal mass and bilateral pleural effusion. Pleural fluid from both hemithorax was yellow and turbid but odorless. Aerobic culture and cytology of pleural fluid were negative. Triglyceride (TG) of both pleural fluids was >110 mg/dL with the ratio of cholesterol/triglyceride of pleural fluid <1 supporting chylothorax. The core biopsy analysis was negative. Non-Hodgkin lymphoma was established by open thoracotomy biopsy and immunochemistry examination. Chylothorax prognosis was an improvement which was reduced after chest tube insertion. On the outpatient, the patient plans chemotherapy with R CHOP regimen (Rituximab + Cyclophosphamide, prednisone, doxorubicin, and vincristine).
Discussion: Malignancy is the primary cause of non-traumatic chylothorax and thoracotomy is used to repair the thoracic duct.
Conclusion: Bilateral chylothorax and non-Hodgkin lymphoma were confirmed based on pleural fluid analysis, thoracotomy open biopsy, and immunochemistry examination.

1. Introduction

Bilateral chylothorax accumulates lymphatic fluid in both pleural cavities due to leakage [1,2]. The incidence of chylothorax has been reported in as many as 3 % of cases of pleural effusions [3] and mortality has been reported in as many as 10 % [4]. In addition, the most common cause of chylothorax is lymphoma (70 %) [5]. Bilateral chylothorax is a rare case that is important to report [6]. This study reported an Indonesian female with bilateral chylothorax and mediastinal non-Hodgkin lymphoma based on SCARE 2020 guidelines [7].

2. Case presentation

An Indonesian female, 34 years old, complained of short breathing, cough, and swelling in some areas. The patient had short breathing for 1 month and worsened for 1 week. The patient also coughed up phlegm with white color >1 month. The swelling was found in the neck, breast, and both arms. The patient did not have hypertension, diabetes mellitus, and asthma. Vital signs were unstable, including heart rate (HR) of 122 ×/min and respiratory rate (RR) of 28 ×/min with the nasal cannula of 3 L/min (SO_2 of 98 %). Physical investigation showed neck edema, increased jugular venous pressure and enlarged lymph nodes that were challenging to evaluate. Thorax examination showed asymmetric lung expansion (left lung lagging on expansion), decreased vocal fremitus, dim percussion, and low vesicular sound in 1/3 hemithorax.

Chest X-ray and thorax CT-Scan of the patient showed enhancing solid lesions (size of 12.2 × 7.5 × 12.8 cm) in the anterior mediastinum and bilateral pleural effusions with adjacent compressive atelectasis (Fig. 1). Pleural fluid analysis showed cytology of carcinoma suspect. In contrast, cell block and core biopsy showed no malignancy. Pleural fluid laboratory showed potassium of 2.9 mmol/L, white blood cell of 16.530 × 103/μL, C-reactive protein of 49.31 mg/dL, and LDH of 305 U/L. In addition, when cultured on pleural fluid, no bacteria were found. The patient received oxygen therapy of 3 L/min (nasal cannula), Amiparen of 500 mL/12 h, Furosemide of 20 mg/8 h, Levofloxacin of 750 mg/day, Codeine of 10 mg/4 h, paracetamol of 500 mg/4 h, KSR of 600 mg/8 h,

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https://doi.org/10.1016/j.ijscr.2022.107827
Received 4 October 2022; Received in revised form 28 November 2022; Accepted 28 November 2022
Available online 1 December 2022

and high-calorie high-protein diet of 2100 kcal/day.

In the 3rd week, the fluid pleural color changed to yellowish and the patient underwent a thoracotomy (open lung biopsy). During surgery, fluid pleural effusion production of 700 mL was obtained. The patient was also placed on a thoracic drain with water seal drainage (WSD) on the left and right hemithorax. The results of lipid profile examination in pleural fluid showed exudate with Triglyceride (TG) of >110 mg/dL and total cholesterol of <200 mg/dL (right = TG of 261 mg/dL, total cholesterol of 33 mg/dL and left = TG of 252 mg/dL, total cholesterol of 37 mg/dL). Open biopsy analysis showed malignant round cells suspected of non-Hodgkin lymphoma (Fig. 2). In the 5th week, the WSD patient was released because the pleural effusion fluid production was 80 mL. Immunohistochemical analysis showed a high grade of non-Hodgkin B lymphoma cell type (CD20 positive). The patient received regimen R CHOP (Rituximab + Cyclophosphamide, prednisone, doxorubicin, vincristine) and continuous outpatient chemotherapy.

3. Discussion

The clinical features of a chylothorax depend on the lymph fluid's degree of leakage and the etiologic cause's symptoms. Symptoms of chylothorax resemble the clinical symptoms of pleural effusion in general, but because the excreted fluid is lymph fluid that contains protein, fat, electrolytes, and vitamins, malnutrition and electrolyte disturbances can be found. An immunosuppressed state may result from losing immunoglobulins, T lymphocytes, and proteins, along with leakage of lymph fluid in the pleural cavity [1,8]. Clinical symptoms appear in the form of shortness of breath, dry cough, and discomfort in the chest. A chylothorax rarely causes fever and chest pain because lymph fluid does not cause inflammation of the pleura [9,10].

The fluid in the chylothorax is generally milky white and odorless. However, some studies have shown that the appearance of the fluid in the chylothorax can be yellow, cloudy, and hemorrhagic. The chylothorax fluid is milky white due to its high lipid content, whereas in empyema, the cloudy fluid is caused by the accumulation of leukocytes, bacteria, and cellular debris. Centrifugation can be performed to differentiate between the two. Pleural fluid analysis in the chylothorax may be exudate, although transudative chylothorax fluid is found in some cases. The chylothorax also found dominant lymphocytes, protein 2.2–6 g/dL, high triglycerides (>110 mg/dL), and low LDH (65–220 mg/dL) compared to serum. Some features suggest a characteristic of transudative fluid, dominant neutrophil, or high LDH. This does not rule out the diagnosis of chylothorax, but the researcher must look for other additional causes that cause the fluid leak. The gold standard for diagnosing a chylothorax is the presence of chylomicrons in the pleural fluid on lipoprotein analysis. It is specific for the chylothorax because chylomicrons are not present anywhere other than the lymph fluid under normal conditions [11,12].

Chylothorax generally occurs in the left hemithorax due to its anatomic location. The thoracic duct crosses in the area between the 7th cervical and 3rd thoracic vertebrae from right to left posterior, ending at the left venous angle. Anatomical variations can occur in several individuals but are not numerous [13]. A chylothorax occurs on the right side of the obstruction, which involves the lower thoracic duct and on the left side of the obstruction, it involves the upper thoracic duct [14,15]. The etiology of chylothorax can be divided into 3 major groups, namely traumatic, non-traumatic, and idiopathic. Traumatic causes are divided into 2, namely iatrogenic/surgical (25–50 % of the traumatic group) and non-iatrogenic. Non-traumatic chylothorax can be caused by obstruction of the lymphatic channels, disease of the lymphatic pathways (e.g., lymphangioleiomyomatosis, yellow nail syndrome, Gorham-Stout syndrome, Noonan syndrome), increased production of lymph fluid (e.g., portal hypertension, cirrhosis of the liver), changes in the composition of lymph fluid ("sludging" in chronic lymphocytic leukemia), chest irradiation. The most common cause of non-traumatic chylothorax is malignancy and lymphoma is the most common cause of malignancy-causing chylothorax (70 %) [13,14].

There are two mechanisms for the occurrence of chylothorax due to lymphoma. The first is the thoracic duct rupture, which may be due to infiltration of malignancy, causing the thoracic duct to become rigid and prone to rupture. The second mechanism is excessive pressure on the thoracic duct. This results in the backflow of lymph fluid into the

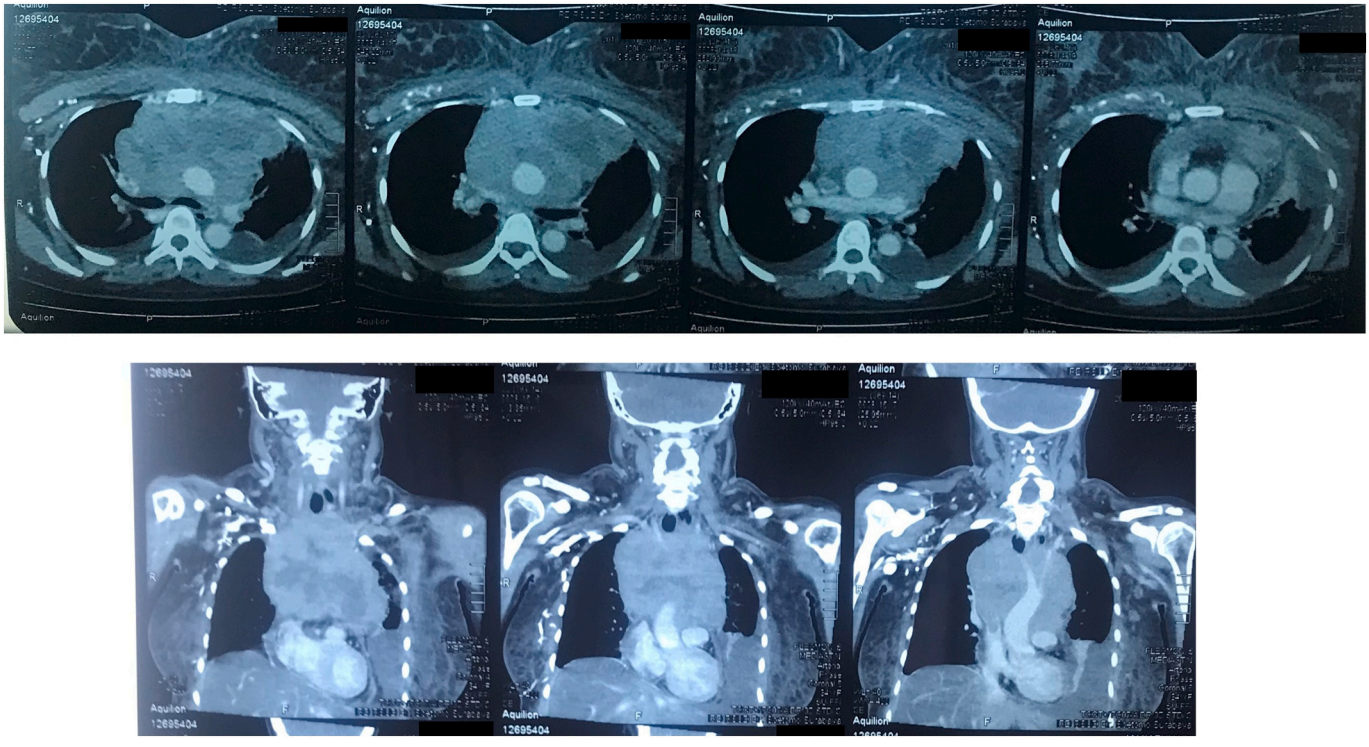

Fig. 1. CT-Scan showed enhancing solid lesion in the mediastinal anterior-posterior.

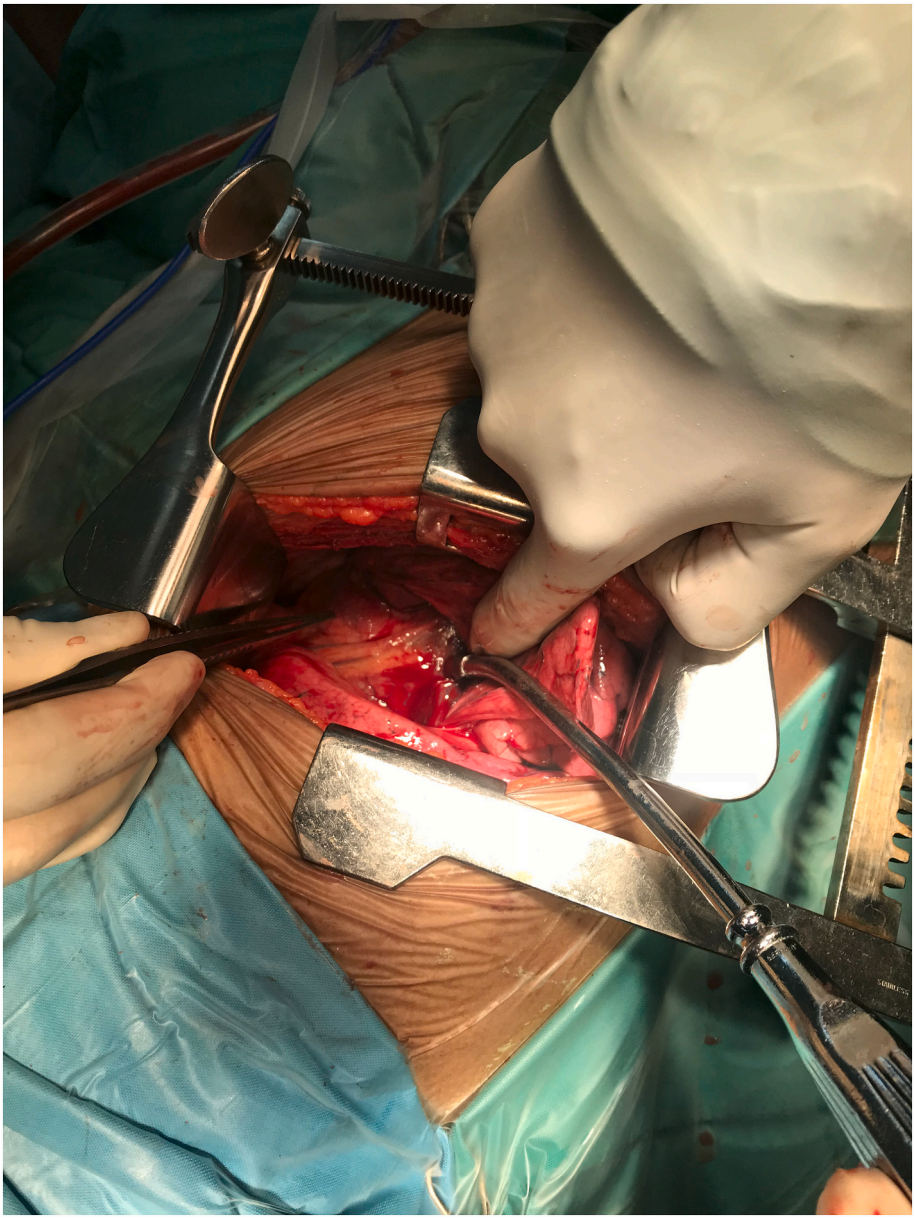

Fig. 2. An open thoracotomy biopsy showed tumor tissue adhering to the left lung and pericardium.

parietal pleura through the lymphatics and then into the pleural cavity. This pressure also forms collateral canals that dilate and open the pleural cavity [16,17]. Another theory states that a large number of lymphocytes and protein material in the lymph fluid in patients with lymphoma and leukemia can cause a high viscosity of the lymph fluid. The high viscosity of the lymph fluid causes tremendous pressure on the walls of the lymph vessels and this high intraluminal pressure causes distension and fragility of the lymph vessels. Minor trauma such as a loud cough or sneeze can cause microrupture of the fragile vessel walls resulting in leakage of lymph fluid into the pleural cavity [18,19].

4. Conclusion

Bilateral chylothorax is a rare condition and the appearance can resemble an empyema. Malignancy is the primary cause of non-traumatic chylothorax, which is mainly lymphoma. Non-Hodgkin lymphoma has a higher incidence of chylothorax than Hodgkin lymphoma. Bilateral chylothorax and non-Hodgkin lymphoma were confirmed based on pleural fluid analysis, open thoracotomy biopsy, and immunochemistry examination.

Source of funding

None.

Ethical approval

Not applicable.

Consent

Written informed consent was obtained from the patient or guardian for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

All authors contributed toward data analysis, drafting and revising the paper, gave final approval of the version to be published and agree to be accountable for all aspects of the work.

Registration of research studies

Not applicable.

Guarantor

Winariani Koesoemoprodjo is the person in charge of the publication of our manuscript.

Declaration of competing interest

Sisilia Yolanda Wijaya and Winariani Koesoemoprodjo declare that they have no conflict of interest.

Acknowledgement

We would like thanks to our editor, “Fis Citra Ariyanto”.

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