World Journal of Advanced Research and Reviews
eISSN: 2581-9615 CODEN (USA): WJARAI
Cross Ref DOI: 10.30574/wjarr
Journal homepage: https://wjarr.com/

(CASE REPORT)

Bilateral chylothorax revealing follicular lymphoma: A case report

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World Journal of Advanced Research and Reviews, 2026, 31(01), 284–290

Publication history: Received on 28 May 2026; revised on 04 July 2026; accepted on 06 July 2026

Article DOI: https://doi.org/10.30574/wjarr.2026.31.1.1846

Abstract

Introduction: Chylothorax is a rare condition resulting from the accumulation of chyle within the pleural space secondary to disruption, obstruction, or rupture of the thoracic duct. Bilateral chylothorax represents a particularly rare clinical presentation and can compromise the patient's functional and nutritional prognosis.

Case Presentation: We report the case of a 61-year-old man with a history of type 2 diabetes mellitus and active chronic smoking who was admitted for progressively worsening dyspnea associated with lower-limb edema and abdominal distension, without any other respiratory or systemic symptoms. The condition evolved in a context of apyrexia and preserved general health status.

Clinical examination revealed a hemodynamically stable patient with no signs of respiratory distress. Bilateral pitting edema of the lower extremities and signs of bilateral pleural effusion syndrome were noted. Chest radiography demonstrated bilateral pleural effusions of moderate volume, more pronounced on the left side.

Diagnostic thoracentesis yielded a milky pleural fluid. Biochemical analysis of the pleural fluid revealed a protein level of 4.7 g/dL, lactate dehydrogenase (LDH) of 102 U/L, triglycerides of 246 mg/dL, and cholesterol of 75 mg/dL. A diagnosis of chylothorax was therefore established, and pleural drainage was performed.

As part of the etiological workup, thoraco-abdomino-pelvic computed tomography and positron emission tomography-computed tomography were performed. These investigations revealed cervical, mediastinal, and abdominal lymphadenopathy suspicious for malignancy, as well as a large retroperitoneal abdominal mass causing compression of the inferior vena cava, associated with bilateral pleural effusions predominantly on the left side.

A right inguinal lymph node biopsy was subsequently performed. Histopathological examination, supplemented by immunohistochemical analysis, established the diagnosis of low-grade follicular lymphoma.

The clinical course was marked by recurrence of the chylous pleural effusion despite initial drainage, requiring multidisciplinary management and initiation of specific treatment for the underlying hematologic malignancy.

Conclusion: Bilateral chylothorax remains a rare clinical entity, the diagnosis of which relies on pleural fluid analysis. Early management, together with treatment of the underlying etiology, is crucial in determining the prognosis.

Keywords: Chylothorax; Pleural effusion; Follicular lymphoma; Retroperitoneal lymphadenopathy; Thoracic duct

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1. Introduction

Chylothorax is a rare clinical condition characterized by the accumulation of chyle within the pleural cavity as a result of injury, obstruction, or rupture of the thoracic duct, the main conduit responsible for chyle transport. (1,2) Although it accounts for only a small proportion of pleural effusions, chylothorax may be associated with significant morbidity and mortality owing to its respiratory, nutritional, and immunological consequences. (3,4)

The etiologies of chylothorax are broadly classified into traumatic and non-traumatic causes. Traumatic chylothorax most commonly occurs following thoracic, cardiovascular, or esophageal surgery, and less frequently after blunt or penetrating thoracic trauma. (5) Non-traumatic causes are predominantly related to malignancies, particularly lymphomas, which represent the leading cause of non-traumatic chylothorax in adults. (1)

The diagnosis relies primarily on pleural fluid analysis. Elevated triglyceride levels (>110 mg/dL) together with reduced cholesterol levels (<200 mg/dL) constitute the principal diagnostic criteria routinely used in clinical practice. (3,6)

The management of chylothorax is multifaceted. Conservative measures, including dietary modifications and pharmacological therapies, are generally considered first-line treatment. Drainage of the chylous effusion may be required to alleviate symptoms and improve respiratory function. In cases refractory to conservative management, interventional procedures such as thoracic duct ligation or embolization may be considered. (3,7,8)

Given the rarity of this clinical entity, each reported case contributes valuable insights into its pathophysiology, diagnostic approach, and therapeutic management. We report an original case of bilateral chylothorax revealing an underlying follicular lymphoma, highlighting the atypical nature of its presentation and the diagnostic challenges associated with this condition.

2. Case Presentation

We report the case of a 61-year-old man with a history of type 2 diabetes mellitus and active chronic smoking who was admitted for progressively worsening dyspnea associated with lower-limb edema and abdominal distension. The clinical presentation evolved in an afebrile setting, without deterioration of the general condition or other significant respiratory or systemic manifestations.

On physical examination, the patient was hemodynamically stable and showed no signs of respiratory distress. Bilateral pitting edema of the lower extremities and clinical signs consistent with bilateral pleural effusion were noted. Chest radiography revealed bilateral pleural effusions of moderate volume, predominantly on the left side.

Diagnostic thoracentesis yielded a milky pleural fluid. Biochemical analysis demonstrated a protein concentration of 4.7 g/dL, lactate dehydrogenase (LDH) level of 102 IU/L, triglyceride concentration of 246 mg/dL, and cholesterol level of 75 mg/dL. These findings were consistent with the diagnosis of chylothorax, prompting the placement of a therapeutic pleural drain.

The etiological workup, including contrast-enhanced thoraco-abdomino-pelvic computed tomography (CT) and positron emission tomography/computed tomography (PET/CT), revealed multiple hypermetabolic cervical, mediastinal, and abdominal lymphadenopathies suggestive of malignancy. These findings were associated with a large retroperitoneal abdominal mass encasing major vascular structures and exerting compressive effects on the inferior vena cava. Bilateral pleural effusions, more pronounced on the left side, were also identified.

A right inguinal lymph node biopsy was subsequently performed. Histopathological examination, supported by immunohistochemical analysis, established the diagnosis of low-grade follicular lymphoma.

The clinical course was marked by recurrence of the chylous pleural effusion despite initial pleural drainage, necessitating a multidisciplinary management approach and the initiation of specific therapy targeting the underlying hematologic malignancy.

3. Discussion

Chylothorax is a rare complication resulting from disruption of thoracic lymphatic drainage due to rupture, obstruction, or infiltration of the thoracic duct. Although it accounts for less than 3% of all pleural effusions, it is associated with

significant morbidity because of the continuous loss of lipids, proteins, immunoglobulins, electrolytes, and lymphocytes, which may lead to severe malnutrition, immunosuppression, and an increased risk of opportunistic infections (1–4). The severity of this condition depends on both the volume of chyle leakage and the underlying etiology, with malignant causes generally associated with a less favorable prognosis (1,3).

In adults, the causes of chylothorax are traditionally classified into traumatic and non-traumatic categories. Thoracic surgical procedures currently represent the leading traumatic cause, whereas malignancies account for the majority of non-traumatic cases (1,2,5). Among these, lymphomas are responsible for nearly 70% of reported cases in the major clinical series (5,9). From a pathophysiological standpoint, the most common mechanism involves thoracic duct involvement through extrinsic compression, direct infiltration, or obstruction by mediastinal or retroperitoneal lymphadenopathy. This results in elevated lymphatic pressure and subsequent leakage of chyle into the pleural cavity (2,3).

Follicular lymphoma is generally an indolent B-cell lymphoproliferative disorder characterized by the neoplastic transformation of germinal center B lymphocytes. Its clinical course is typically slow, although it remains incurable in advanced stages. It commonly presents with diffuse lymphadenopathy, bone marrow involvement, and splenomegaly, whereas extranodal manifestations are less frequent (10,11).

In our case, the occurrence of bilateral chylothorax represents a particularly unusual presentation. The laterality of chylous effusions is closely related to the anatomical site of thoracic duct involvement. Obstruction below the fifth thoracic vertebra most often results in a right-sided pleural effusion, whereas lesions located above this level generally lead to a left-sided chylothorax because of the anatomical course of the thoracic duct within the posterior mediastinum. Bilateral forms remain rare and are usually observed in the setting of major central thoracic duct obstruction or extensive lymphatic infiltration causing a global impairment of lymphatic return to the venous circulation (1,12). In lymphoma-associated chylothorax, several authors have emphasized that bulky mediastinal or retroperitoneal lymphadenopathy may compress or infiltrate the thoracic duct, leading to increased lymphatic pressure and chyle leakage into the pleural cavities (1,5,12). In our patient, the presence of a large retroperitoneal mass encasing the vascular structures, together with diffuse hypermetabolic lymphadenopathy demonstrated on PET-CT, most likely represented the principal pathophysiological mechanism responsible for impaired lymphatic drainage and the development of bilateral chylothorax.

The diagnosis of chylothorax relies primarily on biochemical analysis of pleural fluid. Although a milky appearance is classically considered characteristic, it is observed in only approximately half of cases, making systematic assessment of lipid composition essential. Diagnostic criteria include a pleural fluid triglyceride concentration greater than 110 mg/dL and a cholesterol level below 200 mg/dL, a profile highly suggestive of chylothorax (1,3,6). In our case, pleural fluid analysis revealed triglyceride and cholesterol levels of 246 mg/dL and 75 mg/dL, respectively, findings consistent with this diagnosis. In cases with intermediate or discordant biochemical results, demonstration of chylomicrons by lipoprotein electrophoresis remains the gold standard for confirming the chylous nature of the effusion (1,5).

The principal differential diagnosis of chylothorax is pseudochylothorax, which occurs predominantly in long-standing chronic pleural effusions, particularly in the context of pleural tuberculosis or rheumatoid arthritis. Unlike chylothorax, pseudochylothorax exhibits a biochemical profile characterized by elevated cholesterol levels and low triglyceride concentrations. This composition results from the progressive accumulation of cholesterol crystals within the pleural fluid during the prolonged course of the effusion (13,14).

Imaging plays a central role in the diagnostic workup of chylothorax by facilitating identification of the underlying etiology. Contrast-enhanced thoraco-abdomino-pelvic computed tomography (CT) is the first-line imaging modality for detecting lymphadenopathy, mediastinal or retroperitoneal masses, signs of thoracic duct compression or invasion, and for assessing disease extent (1,5). In lymphoma patients, positron emission tomography-computed tomography (PET-CT) has become a reference imaging modality for initial staging, disease assessment, and prognostic evaluation (15,16). In our patient, PET-CT demonstrated multiple hypermetabolic lymph nodes associated with a large retroperitoneal mass, strongly suggesting an underlying hematologic malignancy. Definitive diagnosis was established by histopathological examination of a lymph node biopsy, which confirmed low-grade follicular lymphoma.

Management of chylothorax requires a multidisciplinary approach combining symptomatic treatment, nutritional support, and therapy directed at the underlying cause. Pleural drainage is indicated in patients with large or symptomatic effusions to relieve dyspnea and improve respiratory function (1,3). Nutritional management is a cornerstone of conservative treatment and consists of a diet low in long-chain triglycerides and enriched with medium-

chain triglycerides, thereby reducing intestinal lymphatic flow and chyle production. Somatostatin or octreotide may also be considered to decrease chyle output (1,3,7).

However, the effectiveness of conservative measures remains limited when the underlying cause persists. In refractory or recurrent chylothorax, interventional procedures such as percutaneous thoracic duct embolization or surgical thoracic duct ligation may be considered (17,18).

In lymphoma-associated chylothorax, control of the underlying hematologic malignancy is essential for achieving sustained resolution of the effusion. Several studies have shown that an effective response to systemic lymphoma treatment is usually associated with a reduction in chyle flow and progressive resolution of pleural effusions (1,5).

Our case illustrates an uncommon presentation of follicular lymphoma revealed by bilateral chylothorax. It highlights the importance of systematically investigating an underlying hematologic malignancy in all cases of non-traumatic chylothorax, particularly when bilateral effusions or deep lymphadenopathy are present. Early diagnostic evaluation combining pleural fluid analysis, imaging studies, and histological confirmation is crucial for guiding appropriate treatment and optimizing patient outcomes.

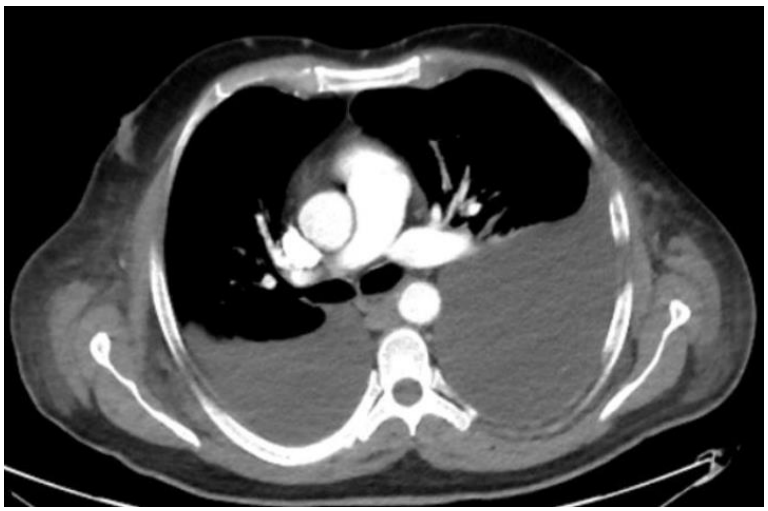

Figure 1 Contrast-enhanced thoracic CT scan showing bilateral pleural effusions, predominantly on the left side.

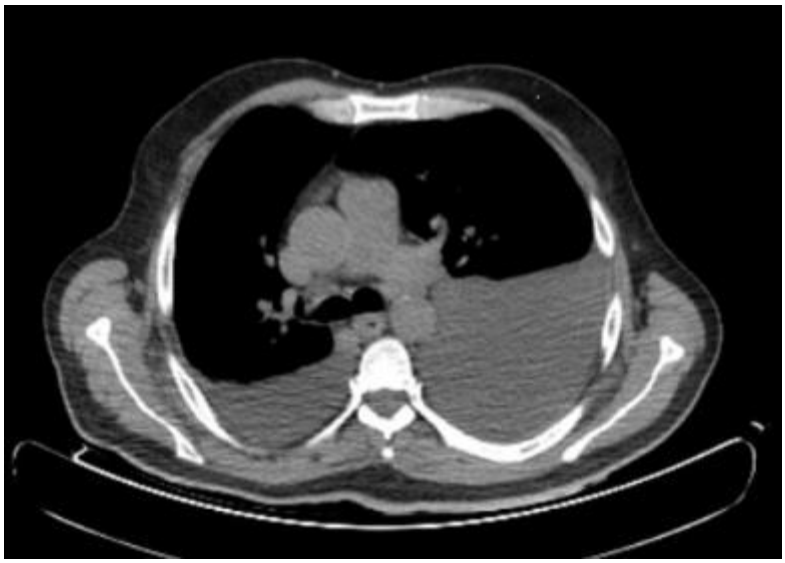

Figure 2 Axial thoracic CT image demonstrating bilateral pleural effusions with compressive adjacent lung atelectasis.

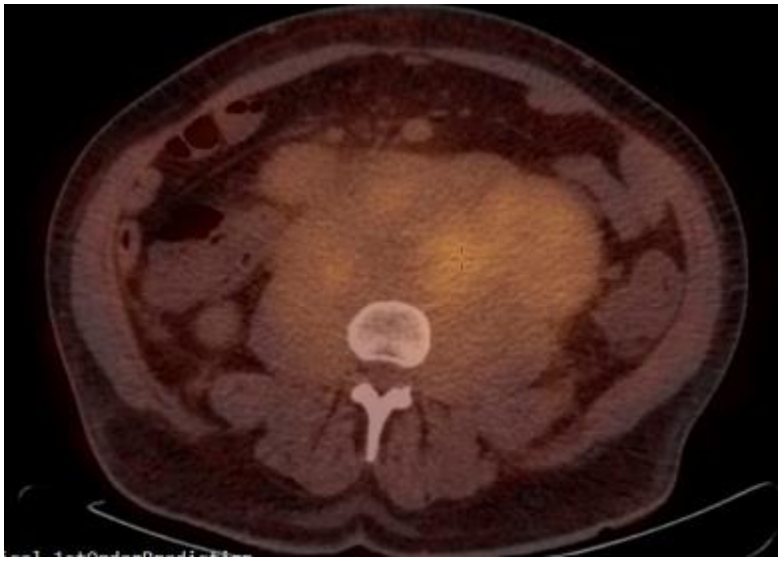

Figure 3 PET/CT image showing a large hypermetabolic retroperitoneal mass encasing major vascular structure.

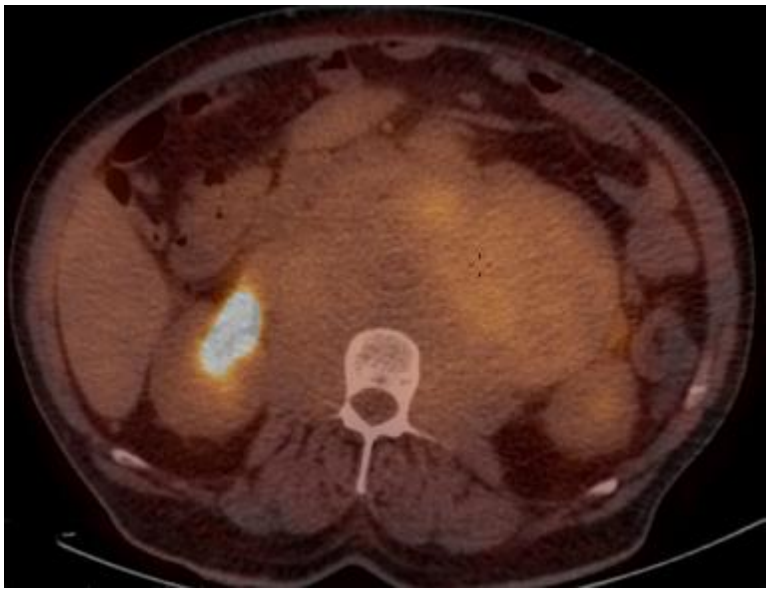

Figure 4 PET/CT image confirming intense FDG uptake within the retroperitoneal mass, suggestive of lymphomatous involvement.

4. Conclusion

Chylothorax is a rare but potentially serious condition that requires prompt diagnosis because of its significant respiratory, nutritional, and immunological consequences. In adults, lymphomas represent the leading cause of non-traumatic chylothorax and should be systematically investigated during the etiological workup, particularly in cases of bilateral involvement. The present case illustrates an atypical presentation of follicular lymphoma, revealed by bilateral chylothorax, most likely resulting from diffuse lymphatic obstruction secondary to mediastinal and retroperitoneal lymphadenopathy. It also highlights the value of a comprehensive diagnostic approach combining biochemical analysis of pleural fluid, imaging studies, and histopathological confirmation. Ultimately, prognosis depends on a multidisciplinary management strategy that integrates symptomatic treatment of the chylous effusion with disease-specific therapy for the underlying hematologic malignancy, the latter being the only intervention capable of achieving sustained resolution of the lymphatic leak**.**

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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