

Neck mass and bilateral pleural effusions in a 53-year-old female

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Abstract

Chylothorax refers to the accumulation of chyle in the pleural cavity and is a rare cause of pleural effusion, especially bilaterally.

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In clinical practice, the presence of milky fluid in the pleural cavity raises suspicion for chylothorax. The most common cause is trauma, iatrogenic or non, resulting from thoracic duct injury, which transports chyle from the lymphatic system into the bloodstream. Here, we present the case of a 53-year-old female who was referred to our hospital with bilateral pleural effusions and a left supraclavicular mass. Diagnostic investigations excluded nontraumatic causes of chylothorax. The potential diagnosis was traumatic chylothorax, a diagnosis of exclusion, as it appeared after muscle strain. This condition was resolved with a fat-free diet and repose without any relapse.

Introduction

Chylothorax is a rare cause of pleural effusion and a common complication of injury to the thoracic duct. The thoracic duct facilitates the transport of chyle from the intra-abdominal lymphatic system to the left subclavian and jugular veins, where it then enters the bloodstream [1]. In chylous pleural effusions, the main mechanism may be a defect in the intrinsic lymphatic mechanism (lymphatic smooth muscle cells and valves) involving impaired lymph valves that permit lymph reflux in the pleural cavity [2]. Furthermore, a mediastinal, cervical, or supraclavicular mass from any cause can be responsible for chylothorax through extrinsic compression or invasion of the thoracic duct [3]. Chylothorax is unilateral in 84% of cases, with 50-60% of all cases being right-sided [4]. Common symptoms include chest pain, dyspnea, orthopnea, and nonproductive cough. Significant loss of immunoglobulins, T lymphocytes, and proteins in the pleural space results in immunosuppression, predisposing patients to opportunistic infections [5]. Chylothorax is classified as traumatic or non-traumatic. The most common cause is trauma, and it is estimated to be up to 40% of all causes. Whether iatrogenic, due to surgical intervention, high-energy trauma, vomiting, or cough, the thoracic duct is damaged [1]. The most common non-traumatic cause is malignancy, especially lymphomas (non-Hodgkin's > Hodgkin's), accounting for approximately 70% of all cases. Idiopathic chylothorax indicates 5-10% of cases [6]. Less common causes are lymphangioliomyomatosis, yellow-nail syndrome, sarcoidosis, tuberculosis, or sarcoma Kaposi. Computed tomography (CT) of the chest, abdomen, and pelvis is recommended to identify sites of traumatic lymphatic injuries, compressive mediastinal or abdominal lymphadenopathy, ascites, or malignant lesions [4]. Magnetic resonance imaging (MRI) lymphography is a useful tool for detecting lymphatic leaks in traumatic cases, allowing the guidance of therapeutic management [3].

Case Report

A 53-year-old Caucasian orthopedics nurse with no remarkable past medical history was referred to our tertiary hospital for

further evaluation of bilateral pleural effusions. She reported the sudden onset of a left supraclavicular mass (Figure 1A) within 48 hours and progressive right-sided pleuritic pain within 24 hours of her admission. There was no remarkable history of trauma, injury, or recent travels, but she mentioned a recent heavy working schedule because of overtime and care of overweight patients the previous days. Radiological imaging with chest X-ray (Figure 2A) confirmed bilateral pleural effusions. A CT scan of the neck, thorax and abdomen confirmed the pleural effusions (Figure 2B) and also revealed a vague inflammatory area at the left side of the neck (Figure 2C) without significant abnormalities in the mediastinum, abdomen, or pelvis.

On presentation, the patient was afebrile and hemodynamically stable. She had an arterial blood pressure of 114/75 mmHg and a heart rate of 90 beats/minute. Peripheral lymph nodes were not palpable, except for the small swelling over the left clavicle with expansion on the left side of the neck. On auscultation, diminished lung



Figure 1. Palpable painless mass over the left clavicle (red arrow).



Figure 2. Bilateral pleural effusions on admission chest-x-ray (A) and on axial image of the thorax in the contrast-enhanced CT (red arrows) (B) and reveal of the vague inflammatory area at the left side of the neck on the coronal image (red arrowheads) (C).

sounds were found in both lung bases without any additional lung sounds. Heart sounds were clear during the examination and there was no evidence of peripheral edema, clubbing, or other abnormalities. The rest of the physical examination was unremarkable.

Laboratory investigations showed mild leukopenia with a white blood cell count of $3.6 \times 10^3/\text{mL}$ and mild anemia with a hemoglobin of 11.4 g/dL. She had a mildly elevated C-reactive protein (7.9 mg/dL, normal values: 0.01-0.5 mg/dL) and serum amyloid A (8.37 mg/dL, normal values: 0.00-0.68). Her basic metabolic panel did not show any significant electrolyte or lipid abnormalities. She had normal triglycerides and cholesterol levels (75.8 mg/dL and 153.3 mg/dL, respectively; normal values: 0.000-200.000). Renal and hepatic functions, likewise serum immunoglobulins, were normal. Serum autoantibodies and thyroid function tests were also within normal range. Hepatitis B virus, hepatitis C virus, and human immunodeficiency virus serologic results were negative. The treponema test was also negative. A Mantoux test was measured at 0 mm, and the peripheral blood smear did not indicate any lymphoproliferative disorder.

Diagnostic thoracentesis of both right and left pleural effusions revealed milky/white-appearing fluid (Figure 3), which was exudative according to Light's criteria. In particular, the fluid total protein (3.8 g/dL)/serum total protein (5.94 g/dL) ratio was 0.63 (>0.5), and the fluid lactate dehydrogenase (193

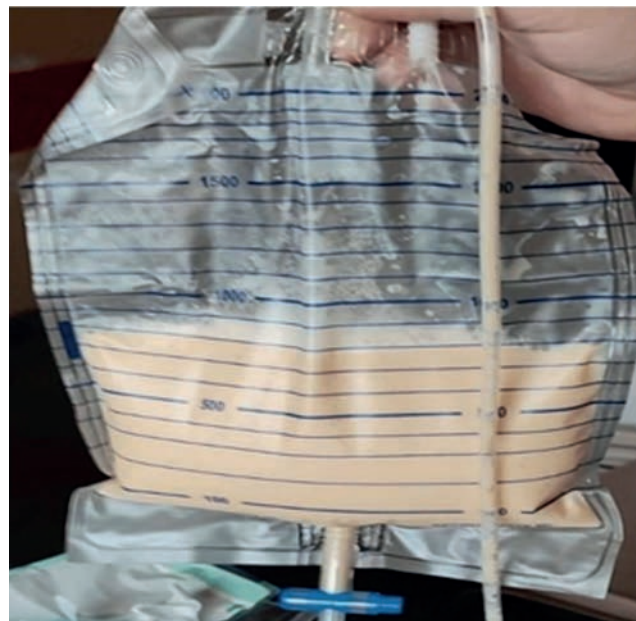


Figure 3. Milky appearance of the pleural fluid.

IU/L)/serum lactate dehydrogenase (203 IU/L) ratio was 0.95 (>0.6). Triglyceride levels were 4013 mg/dL and 2394 mg/dL (>100 mg/dL) on the right and left pleural fluid, respectively, and the cholesterol levels were 140 mg/dL on both sides. The microscopic evaluation did not indicate any cholesterol crystals but revealed a predominance of polymorphonuclear cells. Three cytologic examinations of the pleural effusion, immunophenotyping and polymerase chain reaction for acid-resistant bacteria and acid-fast stain were negative.

An MRI scan of the neck and thorax that demonstrated increased T1 signal intensity at the left supraclavicular area without any anatomic disruptions of blood vessels or lymphatic system. Afterward, a therapeutic thoracentesis was performed, with which the pleuritic fluid dried up and the pleurodynia receded. As for the neck mass, the otolaryngologist's consultation suggested antibiotic treatment, and it improved with automatic remission, so no further diagnostic workup was needed. Consequently, a dietitian consultation was performed, and an appropriate daily diet plan was suggested; on her follow-up after two weeks of repose, she presented a new chest X-ray with complete resolution of the pleural effusions, and she remained asymptomatic.

Discussion

This case describes a potential non-iatrogenic traumatic bilateral chylothorax which resolved with rest and a diet poor in triglycerides and cholesterol. In clinical practice, the presence of milky fluid in the pleural cavity raises suspicion of chylothorax. Regarding the pleural effusions, due to increased levels of triglycerides >100 mg/dL, triglycerides/cholesterol ratio >1, and the absence of cholesterol crystals, the diagnosis of bilateral chylothorax was made [4]. Her excluded medical history was unremarkable, and diagnostic studies, the nontraumatic causes of chylothorax. Subsequently, she was treated as a case of traumatic chylothorax and underwent therapeutic thoracentesis of her effusions, which gradually dried up. Despite the persistent questions about potential trauma in the last few weeks, there was nothing from her medical history that could explain it. Nevertheless, she works as a nurse in an orthopedics department, which involves physically demanding tasks daily and possibly involuntary trauma, which probably was a muscle stretch in the left supraclavicular area that irritated the lymph vessels and created the palpable mass. Consequently, traumatic chylothorax was suspected as a diagnosis of exclusion [7], as it appeared after muscle stretch and receded with an appropriate diet and repose without any relapse. A conservative approach consisting of a fat-free diet was initiated, and following the therapeutic thoracentesis, the effusions dried up. Following 5 days of hospitalization, the patient was discharged in good condition. In her follow-up, her pleural effusions had resolved completely, and her

bloodwork revealed only a mild increase in cholesterol levels (253 mg/dL, normal values: <200 mg/dL), prompting a referral to a specialist for further evaluation.

Currently, there are no official, evidence-based guidelines for the management of chylothorax, so a multidisciplinary approach is required [4]. It includes treatment of the underlying condition and whether conservative or surgical management is appropriate. Treatment of the underlying condition leads to an improvement in the chylothorax or the disease burden (*e.g.*, lymphoma) without necessarily improving the chylothorax. Conservative treatment initially involves replacing the nutrients lost in the chyle and draining large chylothoraces using a chest tube [5]. For chylothorax that has a high output via chest tube (>1100 mL/24 h), usually traumatic, surgical intervention, such as thoracic duct ligation or embolization, is recommended. For effusions that drain slowly, usually non-traumatic, medical management with dietary modifications and somatostatin analogs is preferred [1].

Conclusions

To conclude, chylothorax is a rare cause of pleural effusion, especially bilaterally. The most common cause is trauma, either iatrogenic or not medically related. Moreover, the diagnosis of traumatic chylothorax can be a diagnosis of exclusion when non-traumatic causes can be ruled out. As lymphomas are the most common cause of nontraumatic chylothorax, they should be included in the differential diagnosis. In the absence of specific guidelines for the management of patients with chylous effusions, a multifaceted approach is suggested.

References

1. Ruiz de Villa A, Spencer S, Sircar S, et al. An unusual case of non-traumatic chylothorax. *Cureus* 2022;14:e32506.
2. Zarogiannis S, Hatzoglou C, Molyvdas PA, Gourgoulianis K. Yellow nail syndrome chylous pleural effusions: defective lymph valves involved? *Chest* 2008;134:1353.
3. Cholet C, Delalandre C, Monnier-Cholley L, Le Pimpec-Barthes. Nontraumatic chylothorax: nonenhanced MR lymphography. *Radiographics* 2020;40:1554-73.
4. Riley LE, Ataya A. Clinical approach and review of causes of a chylothorax. *Respir Med* 2019;157:7-13.
5. McGrath EE, Blades Z, Anderson PB. Chylothorax: aetiology, diagnosis and therapeutic options. *Respir Med* 2010;104:1-8.
6. Beg M, Arif H. Recurrent chylothorax in renal cell carcinoma. *Cureus* 2019;22:e5196.
7. Bottet B, Melki J, Levesque H, et al. Stretching and chylothorax. *Rev Mal Respir* 2019;36:742-6. [Article in French].