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

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Abstract

Chylothorax indicates the accumulation of chyle in the pleural cavity. It is a rare cause of pleural effusion, especially bilaterally. In clinical practice, the presence of milky fluid in the pleural cavity raises the suspicion of chylothorax. The most common cause is trauma, iatrogenic or non, owing to thoracic duct injury, which transports chyle from the lymphatic system into the bloodstream. The case we describe is of a 53-year-old female who was referred to our hospital with bilateral pleural effusions and a left supraclavicular mass. From the diagnostic studies, the nontraumatic causes of chylothorax were excluded. The potential diagnosis was traumatic chylothorax, a diagnosis of exclusion, as it appeared after muscle stretch and receded with a fat-free diet and repose without any relapse.

Key words: trauma, non-Hodgkin's lymphoma, milky fluid, pleural effusion, chylothorax.

Introduction

Chylothorax is a rare cause of pleural effusion and a common complication of injury to the thoracic duct. The thoracic duct helps transport chyle from the intra-abdominal lymphatic system to the left subclavian and jugular veins where it then enters the bloodstream circulation [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 by extrinsic compression or invasion of the thoracic duct [3]. The effusion occurs unilaterally in 84% of cases, with 50-60% of all cases being right-sided [4]. The most common symptoms are chest pain, dyspnea, orthopnea, and nonproductive cough. Significant loss of immunoglobulins, T lymphocytes, and proteins into the pleural space results in immunosuppression, predisposing patients to opportunistic infections [5]. It can be classified as traumatic or non-traumatic. The most common cause is trauma and it is estimated 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.

It is estimated that 5% to 10% of chylothorax cases are idiopathic [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 injury to the lymphatic system, compressive mediastinal or abdominal lymphadenopathy, ascites, or malignant lesions [4]. Magnetic resonance imaging (MRI) lymphography is a useful tool for identifying the site of the lymphatic leak, especially in cases of traumatic chylothorax, 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 from another hospital for further evaluation of bilateral pleural effusions. The patient noticed an acute appearance of a mass at the left supraclavicular area (Figure 1A) within 48 hours and progressive right pleuritic pain 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 overtimes and care of overweight patients the previous days. Radiological imaging with chest-x-ray (Figure 2A) showed bilateral pleural effusions and neck-thoracic-abdomen computed tomography (CT) confirmed the pleural effusions (Figure 2B) and also revealed a vague inflammatory area at the left side of the neck (Figure 2C). No significant abnormalities were detected in the mediastinum, abdomen, or pelvis.

At 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 sounds were found in both lung bases without any additional lung sounds. Heart sounds were normal and clear during the examination. There was no peripheral edema, clubbing, or other abnormalities. The rest of the physical examination was unremarkable. Laboratory results showed mild leukopenia with a white blood cell (WBC) count of $3.6 \times 10^3/\text{mL}$ and mild anemia with a hemoglobin (Hb) of 11.4 g/dL. She had a mildly elevated C-reactive protein (CRP) (7.9 mg/dl, normal values; 0.01-0.5 mg/dl) and Serum Amyloid A (SAA) (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 153.3mg/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 (HBV), hepatitis C virus (HCV), and human immunodeficiency virus (HIV) serologic results were negative. The treponema test was also negative. The Mantoux test was measured at 0mm. Peripheral blood smear did not indicate any lymphoproliferative disorder. A diagnostic thoracentesis was performed on both right and left pleural effusion. The diagnostic thoracentesis 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 IU/l)/serum lactate dehydrogenase (203 IU/l) ratio was 0.95 (>0.6). Triglyceride levels were 4013mg/dl and 2394mg/dl (>100mg/dl) on 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. There was also a predominance of polymorphonuclear cells. Three cytologic examinations of the pleural effusion were negative, and immunophenotyping showed nothing irregular. Polymerase chain reaction (PCR) for acid-resistant bacteria and acid-fast stain were negative. After the initial evaluation, she underwent a magnetic resonance imaging (MRI) scan of the neck and thorax that demonstrated increased T1 signal intensity at the left supraclavicular area without any anatomic disruptions on blood vessels or lymphatic system. Afterwards, 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, and on her follow-up after two weeks of repose, she presented a new chest x-ray with remission of pleural fluid bilaterally.

Discussion

This is a case of a potential non-iatrogenic traumatic bilateral chylothorax in which remission has been observed with rest and a diet poor in triglycerides and cholesterol. In clinical practice, the presence of milky fluid in the pleural cavity raises the 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 previous medical history was unremarkable. From the diagnostic studies, the nontraumatic causes of chylothorax were excluded. 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 supracalvicular area which 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. After five days of hospitalization, she was discharged in good condition. In her follow-up, there was remission in the pleural effusions and her bloodwork revealed only a mild increase in cholesterol levels (253 mg/dl, normal values; <200 mg/dl), so she was recommended visiting an expert.

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. 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/24hr), 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 nontraumatic causes can be ruled out. As lymphomas are the most common cause of nontraumatic chylothorax, they should be included in the differential diagnosis. Nevertheless, as there are no specific guidelines for the management of patients with chylous effusions, a multifaceted approach is suggested.

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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).



Figure 3. Milky appearance of the pleural fluid.