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An Unusual Presentation of Follicular Lymphoma

Presentación no habitual del linfoma folicular

To the Editor:

The presence of chyle in the pleural space defines chylothorax.[1] It is usually generated by an alteration in the pathway of the thoracic duct. Neoplasias represent between 30 and 50% of the cases, and non-Hodgkin's lymphoma (NHL) is the most common pathology.[2] Even in the centers of concentration, chylothorax is an infrequent pathology. We present for consideration the even less frequent case of bilateral chylothorax.

The patient is a 66-year-old man who complained of oppressive pain in the upper hemiabdomen, immediate postprandial fullness, hyporexia and a weight loss of 10 kilograms, all over a period of two months. The subject underwent esophagogastroduodenoscopy, revealing non-sliding hiatal hernia. He received treatment, with no improvement. One month later, he started with dyspnea when climbing stairs (MRC 1), and within 3 weeks he started with dyspnea at rest (MRC 4), at which time he came to our hospital for evaluation. Upon physical examination, bilateral pleural effusion was found (confirmed on chest radiograph) as were 1-cm non-painful adenomegalies located in the submandibular region and in the right axillary region. Thoracocentesis was performed with pleural biopsy. Histopathology and culture analyses, including acid-fast stain, were reported negative. Pleural liquid analysis (pH 7.4, proteins 4.56 g/dl, lactate dehydrogenase 93 UI/l, triglycerides 1.137 mg/dl, cholesterol 86 mg/dl) confirmed chylothorax. In addition to pleural effusion, thoracic computed tomography documented an increase in density (54 Hounsfield units) of lymph node stations 4 (R and L) and 7, the largest diameter being 1 cm (fig. 1). Bronchoscopy, bronchoalveolar lavage and bronchial biopsy were negative for malignancy as well as for the microbiological analysis. Due to the lack of conclusive results, we contemplated mediastinoscopy. Nevertheless, the right axillary adenomegaly was biopsied. The histopathological analysis showed lymphoid follicular hyperplasia (fig. 1) and immunohistochemical analysis was positive for BCL-6, BCL-2, CD 10, CD 20 and cyclin D1, establishing the diagnosis of stage II follicular lymphoma.

Chemotherapy was initiated and, due to pleural effusion relapse, a pleural catheter was inserted and the patient started a diet with branched-chain triglycerides and total parenteral nutrition. He was discharged after two weeks and has completed eight cycles of chemotherapy. Two months later, the patient is stable, with relapse of pleural effusion in 10% of the thoracic area.

The diagnostic challenge of chylothorax lies in the information compiled during the initial evaluation due to the multiple etiological associations.[1] Less than 50% of cases with chylothorax have the milky appearance, therefore it is crucial to identify it to guide the adjacent mechanism and shorten the differential diagnosis.[2] Chylothorax may be the first finding of a lymphoma, although bilateral presentation is exceptional.[3,4] These are typically lymphocytic exudates, unilateral and only 1 in 10 have the milky appearance.[5] The mechanisms by which chylothorax is associated with lymphomas can be: 1) neoplastic pleural infiltration; 2) obstruction of lymphatic nodes at the mediastinal level due to neoplastic infiltration; and 3) tumor obstruction of the thoracic duct. Mortality in non-traumatic chylothorax has decreased and it is currently less than 40%. However, when it is associated with malignancy and bilateral presentation, it is given a poorer prognosis. Morel et al. reported that the presence of pleural effusion and advanced stages of the disease (Ann Arbor III and IV) influenced survival adversely. The average survival time was 111 months.[6]

The interest for presenting this case lies not only in its unusual presentation, but also in its diagnostic difficulties. With the epidemiological and clinical history, the presence of chylothorax indicated the probable existence of a lymphoma, a diagnosis elusive to the initial methods (bronchoscopy, pleural biopsy, FNP).

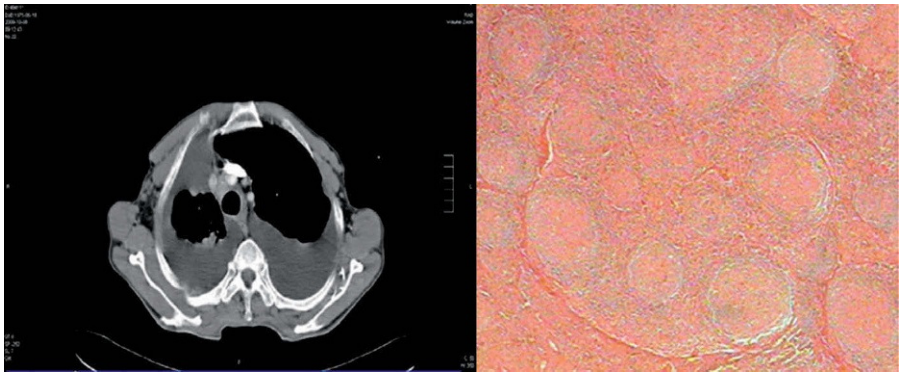

Figure 1. Left panel. Thoracic computed tomography showing an increase in density of lymph node station 4 (R and L). Right panel. Histological cross-section of the axillary lymph node.

Nevertheless, detailed physical examination avoided an invasive approach (mediastinoscopy). The diagnosis of lymphoma was based on immunohistochemical studies and continued to be a challenge for the pathologist. Thus, although not frequently, the presence of bilateral chylothorax can also be associated with lymphoma. The access to a lymph node group can be key in the diagnosis and in the possibility for health of our patients.

Acknowledgements

The authors would like to thank Baltazar Cortés, electronic engineer, for his support in correcting and formatting the images.

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