

Spontaneous bilateral chylothorax: uniform features of a rare condition

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ABSTRACT: A 67-yr-old woman was admitted with dyspnoea, chest and neck pain and swelling of both supraclavicular fossae and the neck. Chest X-ray showed bilateral pleural effusions. Thoracocentesis yielded a milky fluid with a high triglyceride level. Four previously published cases had similar clinical manifestations. The pathophysiology is discussed. *Eur Respir J.*, 1988, 1, 872-873.

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Chylothorax represents about 2% of all pleural effusions in the adult; in 15% of these, no underlying disease is found, and they are classified as spontaneous. In some of the cases, hyperextension of the dorsal spine or a Valsalva manoeuvre may have caused the rupture of a previously altered duct. When the leakage of chyle occurs at a level between the third and sixth dorsal vertebrae, bilateral chylothorax may develop. Only four cases of spontaneous bilateral chylothorax appear to have been reported in the English and Spanish literature [1-4].

Case report

In August 1985, a 67-yr-old woman with an unremarkable medical history, was admitted to our hospital because of dyspnoea and chest pain. She had been in good health until 48 h before admission, when she suddenly felt anterior chest and neck pain, progressive dyspnoea and swelling of both supraclavicular fossae and the neck. There was no history of dorsal hyperextension, surgery or trauma.

On arrival at hospital the patient, was tachypnoeic and cyanotic. Physical examination was consistent with bilateral pleural effusions. Lymph node enlargement, hepatomegaly and splenomegaly were not found. Both supraclavicular fossae and the neck were mildly swollen. Jugular engorgement and facial oedema were absent. Arterial blood gases were: pH=7.48; P_{O_2} =5.8 kPa (44 mmHg); P_{CO_2} =4.5 kPa (34 mmHg); O_2 saturation=81%; HCO_3^- =25.9 mEq.l⁻¹. Routine blood and urine analysis were normal.

Chest X-ray films demonstrated bilateral pleural effusions. Bilateral thoracocentesis yielded a milky fluid with a triglyceride level of 12 g.l⁻¹. Bacteriological

studies were negative and no malignant cells were found. Bone marrow biopsy, bronchoscopy, gastroscopy, oesophagogram and thoracoabdominal computerized tomography (CT) scan were normal.

The patient's condition improved spontaneously, and the pleural effusion had resolved by the tenth day of hospitalization; for that reason, lymphangiography was considered unnecessary. After twentyfour months, the patient was asymptomatic, and a new thoracoabdominal CT scan showed no pathological findings.

Discussion

There is a remarkable uniformity in the clinical manifestations of the reported cases of spontaneous bilateral chylothorax, which has not been previously emphasized. Three cases had chest pain [1-3] and the other had bilateral supraclavicular pain [4], three complained of dyspnoea [1, 3, 4] and all cases presented with supraclavicular and/or neck swelling. In none of the cases was extravasation demonstrated by lymphangiography and all of them resolved spontaneously in less than two weeks.

It has been suggested that pain and dyspnoea are caused by the discharge of the chyle into the pleural space [5], but the significance of the supraclavicular and neck swelling remains obscure. Surprisingly, the latter clinical feature is not usually mentioned when clinical manifestations of chylothorax are described, though it was present in all the reported cases of spontaneous bilateral chylothorax. Its short duration and the absence of jugular engorgement suggest that it may be a manifestation of the mediastinal chyloma that is formed just before the passage of chyle into the pleural cavity. The fact that cervical or supraclavicular fullness is constant in spontaneous

bilateral chylothorax, while it appears to be rare in other groups of chylothorax may indicate that the volume of the chyloma and its rate of formation are greater in spontaneous bilateral chylothorax.

We conclude that spontaneous chylothorax is a rare cause of bilateral pleural effusion, and that it usually requires no aggressive therapeutic measures. In a patient with dyspnoea, chest pain or supraclavicular pain and supraclavicular or neck swelling, the diagnosis of spontaneous chylothorax should be considered.

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RÉSUMÉ: Présentation d'un cas de chylothorax spontané bilatéral chez une femme de 67 ans, admise dans le service pour dyspnée, douleurs thoraciques et cervicales, et gonflement du cou et des deux fosses sus-claviculaires. Nous insistons sur l'uniformité des manifestations cliniques des cas rapportés dans la littérature, et discutons la physiopathologie de cette affection extrêmement rare.