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Development of Idiopathic bilateral Chylothorax in a young female: A rare case report

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Abstract: *Chylothorax is a rare clinical entity characterised by a milky white fluid with increased triglyceride levels and chylomicrons in the pleural cavity. The commonest aetiology is malignancy and trauma. Persistent leak leads to significant loss of essential proteins, immunoglobulins, fat, vitamins, electrolytes and water and needs a prompt treatment. Bilateral idiopathic Chylothorax as observed in the present case is a rare entity.*
Keywords: *Chyle, Thoracic duct, Chylothorax, Triglyceride, Chylomicrons.*

I. Introduction

Chylothorax is a relatively rare cause of a pleural effusion and it occurs when chyle is found in the pleural space. Chyle is lymphatic fluid characterised by high triglyceride and low cholesterol concentration secreted by intestinal cells. It is collected and then transported via the thoracic duct into the circulation. Chylothorax is usually associated with neoplasm or trauma to the thoracic duct. Here we describe a case of idiopathic bilateral Chylothorax observed in a 35 year old female.

II. Case Report

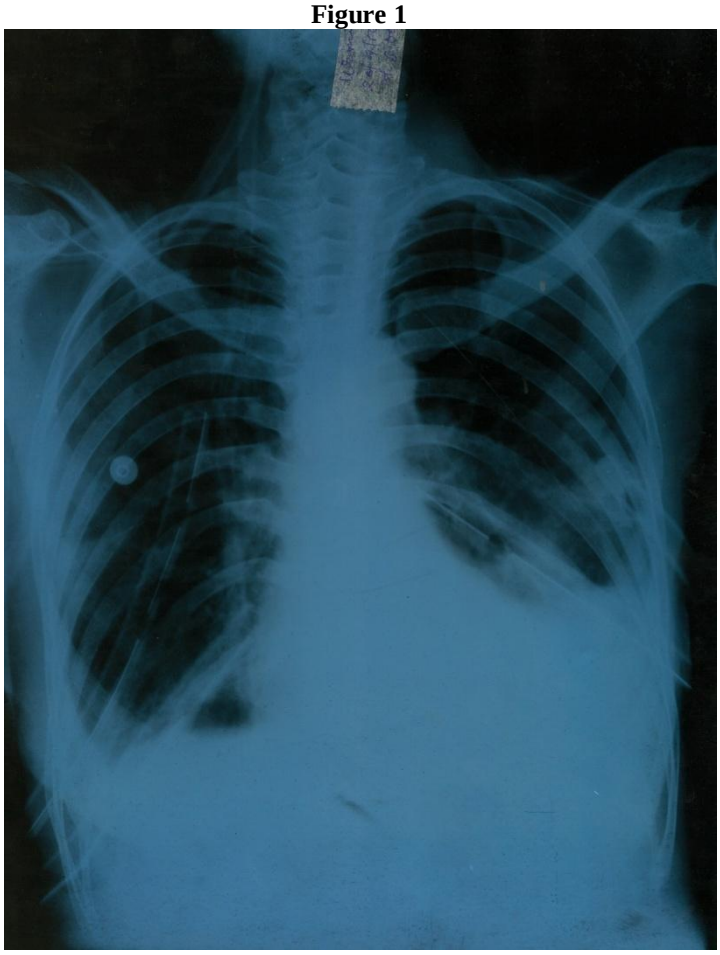
A 35 year old female was admitted to our department with the complaints of breathlessness and chest discomfort from two months. The resting pulse rate was 94/min and the blood pressure was 110/70 mmHg. There was no history of tuberculosis in this patient. The physical examination revealed pallor with no lymph node enlargement. On chest examination, there was stony dull note localised to bilateral infra scapular region and lower axilla.

In blood reports were Hb: 8.6 gm%, Albumin: 3 gm% with normal total and differential leukocyte counts. In chest X-ray there was bilateral pleural effusion (Fig.1) for which bilateral intercostals tube drainage(ICTD) was done. About 1.5 litres and 1.2 litres of milky white pleural fluid was aspirated from right and left chest respectively. Clinical diagnosis of Chylothorax was made. Pleural fluid of both sides was sent for examination separately. That of the right side revealed protein 4.0 g%, sugar 45 mg%, total leukocyte count 3400 cells/mm^3, differential leukocyte count with neutrophils 22, lymphocyte 78, pleural fluid triglyceride 530 mg% and cholesterol 24.6 mg%. Reports of the left side showed protein 4.0 g%, sugar 42 mg%, total leukocyte count 3300 cell/mm^3, differential leukocyte count with neutrophils 35, lymphocyte 65, pleural fluid triglyceride 190 mg% and cholesterol 24 mg% (Table).

Serum triglyceride and serum cholesterol was 72.5 mg% and 72 mg% respectively. The Ziehl-Neelsen stain of the pleural fluid was negative. Serum IgG and IgM was negative for tuberculosis. Mountex test was negative. Ultrasound of abdomen revealed normal study. CT scan chest was done which revealed bilateral pleural effusion with thoracic duct prominent (Fig.2). To confirm the exact side of thoracic duct tear, lymphangiography was planned but her parents refused for further investigations.

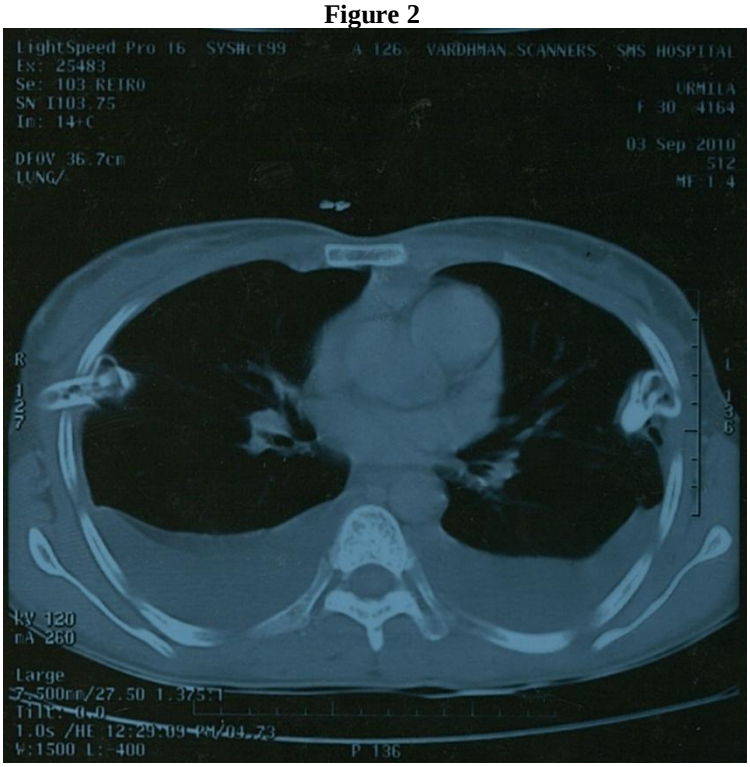
III. Figures And Table

Figure 1



Chest X-ray PA view showing bilateral intercostal tubes with pleural effusion

Figure 2



CECT chest showing bilateral intercostals tubes in-situ with pleural effusion

Table: Charecteristics of pleural fluid on both sides of pleural cavity

Pleural Fluid	Right	Left
Protein	4.0 g%	4.0 g%
Sugar	45.0 mg%	42.0 mg%
TLC	3400 cells/mm^3	3300 cells/mm^3
DLC	P-22,L-78	P-35,L-65
Triglyceride	530 mg%	190 mg%
Chloesterol	24.6 mg%	24 mg%

IV. Discussion

Chylothorax was recognised in the 17th century but it is still a rare entity to be viewed by most physicians as a clinical curiosity. In 1875, H. Quinke described the first traumatic chylothorax and in 1948, R.S. Lampson performed the first thoracic duct ligation. Modified De Meester classification divides chylothorax into congenital, traumatic, neoplastic and miscellaneous category on basis of aetiology[1].

Trauma mostly in form of cardiovascular, pulmonary and oesophageal surgery remains the leading cause of Chylothorax[2].

Another major cause of Chylothorax is malignancy. The most common malignancy to cause Chylothorax is a lymphoma[3] followed by bronchogenic carcinoma[4], and rarely leukaemia.

Congenital Chylothorax is caused possibly because of combination of thoracic duct malformation with sudden elevation of venous pressure and has been reported in conjunction with several syndromes, such as Noonan's and Down's[5].

Causes included in the miscellaneous category are thrombosis of superior vena cava or subclavian vein, cirrhosis, lymphangioleiomyomatosis, Gorham syndrome, Kaposi sarcoma, filirasis, sarcoidosis hypothyroidism, familial lymphedema radiation induced mediastinal fibrosis.

Very few cases of bilateral Chylothorax have been reported in the literature[6]. Chylothorax has no predilection for age and sex. Symptoms of Chylothorax mostly depend upon the amount of fluid in the pleural cavity.

Best way to establish the diagnosis of Chylothorax is to determine the concentration of triglycerides in the pleural fluid. The triglyceride concentration greater than 110 mg/dl (in our case it was 290 mg/dl), a ratio of pleural fluid to serum triglycerides of greater than 1(in our case it was 3.76) and a ratio of pleural fluid to serum cholesterol of less than 1(in our case it was 0.354), confirmed Chylothorax[7]. Bipedal lymphangiography has been recommended to identify the cause and detect the size and site and size of the leak[8].

Primary treatment in the case of Chylothorax should be directed towards the correction of malnutrition and compromised immunologic status[9]. The defect in the thoracic duct often closes spontaneously if the cause is traumatic. In case of severe dyspnoea, the placement of the pleuroperitoneal shunt or chest tube drainage is mandatory[10]. One approach to management of chylothorax has been obliteration of the pleural space, either chemically (tetracycline, talc or povidone-iodine) or surgically[11]. If the Chylothorax persist for more than 4 weeks, consideration should be given to surgical exploration with ligation of the thoracic duct[12].

Diagnosis of Chylothorax, in this case, was established on typical pleural fluid colour with high triglyceride level, high ratio of pleural fluid to serum triglyceride, and low ratio of pleural fluid to serum cholesterol. Patient did not respond to conservative treatment and surgical intervention was planned.

Right Postero lateral Thoracotomy was performed through 5th intercostal space. There were adhesions and generalised oedema, dissection between oesophagus and azygous vein was carried out. Leak of chylous fluid was found between oesophagus and azygous vein which was closed by ligation on both sides of the leak above the diaphragm.

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