

Bilateral Chylothorax with Chylopericardium

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Abstract

A 35-year-old male patient presented with complaints of breathlessness and left-sided chest pain of five days duration. Physical examination revealed a left-sided massive pleural effusion, right-sided moderate pleural effusion and pericardial effusion. Laboratory examination revealed a high triglyceride content in the pleural and pericardial fluids suggestive of chylous pleural effusion and chylopericardium. Further investigations confirmed the diagnosis of malignant lymphoma (thymic large B-cell lymphoma). The report was confirmed with immunohistochemistry markers, by tru-cut biopsy from mediastinal mass. Bilateral chylothorax with chylopericardium is a rare entity and this condition secondary to lymphoma has a poor prognosis. [Indian J Chest Dis Allied Sci 2017;59:147-150]

Key words: Chylothorax, Chylopericardium, Lymphoma.

Introduction

Chylothorax is the accumulation of chyle in the pleural cavity.¹ Chylothorax is suspected when the aspirated pleural fluid appears milky which when kept for sometime forms a creamy layer of chylomicrons. The diagnosis is confirmed by lipoprotein electrophoresis. It will differentiate between chylothorax and pseudochylothorax which contains high content of cholesterol or lecithin globulin complex or both.^{1,2}

Case Report

A 35-year-old male presented with complaints of severe breathlessness, left-sided chest pain to the emergency services of our institute. The patient was asymptomatic five days ago when he developed breathlessness which was acute in onset and was rapidly progressive. The breathlessness was more on lying down on the right side. He also complained of dry cough, dull aching chest pain on the left side which increased on exertion. Patient had no history of diabetes mellitus, hypertension or tuberculosis in the past and had no family history of any illness. He was a farmer by occupation and used to consume alcohol regularly.

On general physical examination bilateral supra-clavicular lymph nodes were present. Respiratory system examination revealed left-sided fullness with trachea shifted towards the right side and decreased left side movements. On percussion, stony dull note was present on whole of the left side. On auscultation breath sounds were absent on the left side.

Chest radiograph (Figure 1) showed a homogeneous opacity in whole of left hemithorax. Total leucocyte count was elevated (21,300 cells/mm³), liver function tests, renal function tests, blood coagulation profile was also normal.



Figure 1. Chest radiograph (postero-anterior view) showing a homogeneous opacity in whole of left hemithorax.

On thoracentesis white milky pleural fluid was aspirated. Intercostal tube drainage was done and three litres of white milky fluid was drained from the left side. Pleural fluid was negative for acid-fast bacilli, (AFB) and malignant cells. Gram staining of pleural fluid did not reveal any organisms and culture was sterile. Pleural fluid total cell count was 750 cells/mm³ with polymorphs 8%, lymphocytes 63% and mesothelial cells 29%. Pleural fluid adenosine deaminase was 58 U/L. Biochemical analysis of the

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pleural fluid revealed glucose 123 mg/dL, protein 2.8 g/dL and cholesterol 35 mg/dL and triglyceride levels were elevated 821 mg/dL suggestive of chylothorax.

Contrast-enhanced computed tomography (CECT) of thorax showed a large mass lesion (12.5cm × 9.7cm × 8.0cm) in anterior and superior mediastinum more towards the left side encasing the trachea and main bronchi with bilateral pleural effusion and mild pericardial effusion (Figures 2). Computed tomography (CT)-guided fine needle aspiration cytology (FNAC) of the mass lesion in the lung as well as supra-clavicular lymph nodes were done but reported as cytology inconclusive. Flexible video bronchoscopy revealed left-sided narrowing of segmental bronchi with mucosal oedema; right side was normal. Bronchial washings were negative for malignant cells, AFB and sterile on culture. The patient was treated symptomatically for a while then the patient was referred to higher centre in last week of November for further evaluation and management on patient's request.

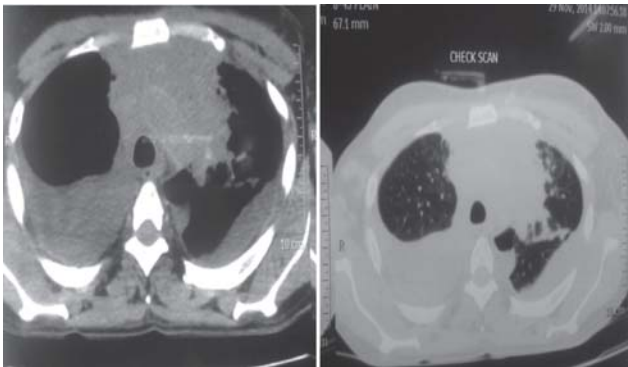


Figure 2. Contrast enhanced computed tomography of thorax showing a large conglomerated mass lesion (12.5cm × 9.7cm × 8cm) in anterior and superior mediastinum more towards left side encasing the trachea and main bronchi with bilateral pleural effusion and mild pericardial effusion.

There the patient was admitted with left-sided ICD *in situ* with fluid draining, chest radiograph was done which revealed moderate effusion on the right side and left sided ICD *in situ* (Figure 3). Diagnostic thoracentesis was done, milky white fluid was aspirated which showed high triglyceride levels (647 mg/dL) suggestive of chylous effusion. At this point, the patient had bilateral chylothorax of unknown aetiology. Thoracoscopy was done on the right side and approximately 1.7 L of milky fluid was aspirated, thoracoscopic pleural biopsy was obtained, intercostals tube drainage was done on the right side and the patient was managed symptomatically.

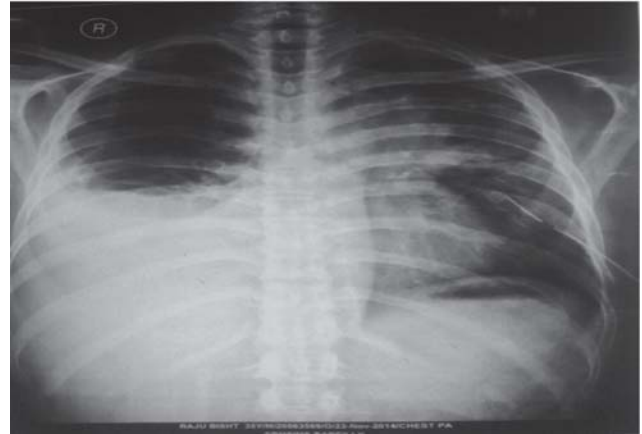
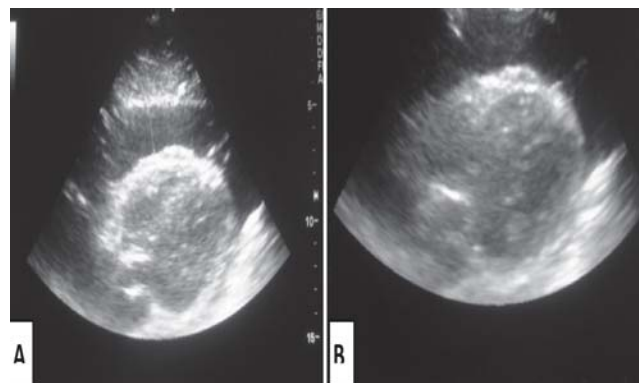


Figure 3. Chest radiograph (postero-anterior view) showing moderate effusion on the right side and left sided intercostal chest tubes *in situ*.

Two days later, the patient developed breathlessness and was shifted to intensive care unit. He was haemodynamically unstable and blood pressure was not recordable. Echocardiography revealed cardiac tamponade (Figures 4), pericardiocentesis was done immediately and around 500 mL of pericardial fluid was aspirated. Laboratory examination of pericardial fluid was suggestive of chylopericardium. Catheter was placed in pericardium and the patient became haemodynamically stable. Thoracoscopic pleural biopsy was negative for malignancy.

Since patient was having bilateral chylothorax with chylopericardium with all biopsy and FNAC done, thus far reported negative for malignancy, CT-guided trucut biopsy was done from the mediastinal mass. Histopathological examination including immunohistochemistry revealed thymic large B-cell lymphoma (Figure 5). We tried pleurodesis after ensuring the fluid free pleural cavity post ICD but failed.

Patient was treated symptomatically, given low fats, high protein diet with triglycerides supplementation. Despite the rapid management and best of care and nutrition patient could not survive and died of shock secondary to respiratory failure.



Figures 4. Echocardiography showing cardiac tamponade.

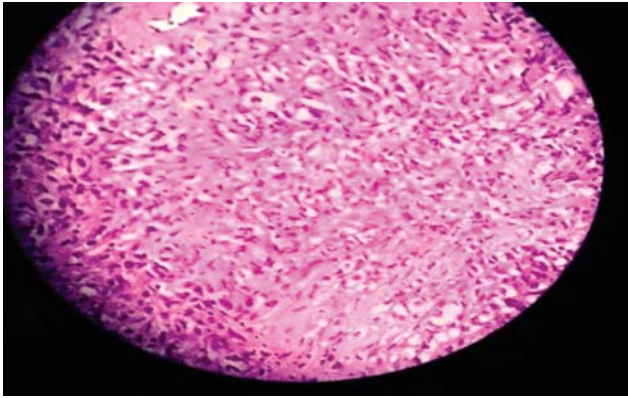


Figure 5. Photomicrograph suggestive of thymic large B-cell lymphoma.

Discussion

Bilateral chylothorax with chylopericardium is an extremely rare condition. Chylothorax can occur as a result of trauma or injury to thoracic duct³ or in oesophageal or cardiothoracic surgery.⁴⁻¹² In thoracic malignancies, chylothorax can occur as a result of blockage to thoracic duct or thoracic lymphatic system.¹³⁻¹⁷ In chylothorax of unknown origin diagnostic difficulties are encountered and malignancy like lymphoma needs to be ruled out.

Chylothorax is usually right sided since most of the thoracic duct is in the right hemithorax. When damage is at the level of aorta, the chyle appears on left.^{18,19} Pseudochylothorax occurs with long-standing fluid having cholesterol and no triglycerides or chylomicrons. In both the conditions the pleural fluid is thick and opaque.²⁰

The causes of chylothorax are divided into four major categories, such as malignancy, traumatic, idiopathic and miscellaneous.² Causes due to trauma are injuries from penetrating wounds surgery of thorax or neck on the left side.⁴⁻¹² Spontaneous chylothorax may occur in lymphoma or bronchogenic carcinoma.^{14,15} Sometime rare causes, like congenital chylothorax, aortic aneurysm, filariasis, subclavian vein thrombosis and thoracic duct tumours, can result in chylothorax.^{21,22} The diagnosis of chylothorax is confirmed by lipoprotein analysis of the fluid. The total lipid varies from 4 to 40 g/L and it has high triglyceride content and low cholesterol concentration. Lipoprotein electrophoresis will show the chylomicron band² in chylothorax which typically contains erythrocytes $05-06 \times 10^9/L$ and lymphocytes $4-6.0 \times 10^9/L$ and a high protein content (>30 g/L).^{16,23}

Diagnostic dilemma remained in our case in spite of all investigations, like thorascopic pleural biopsy, lymphnode FNAC, bronchoscopy with BAL. In our

case diagnosis was established to be thymic B-cell lymphoma on CT-guided tru-cut biopsy from the mass lesion.

Management of chylous effusion comprises maintenance of nutrition, the reduction of flow in the thoracic duct, minimise the chyle formation, parental nutrition, high protein diet with dietary fats replaced with triglycerides or both and bed rest.²⁴⁻²⁷ Symptomatic management includes reducing breathlessness by repeated aspiration of chylous pleural fluid for 4-5 weeks or intercostal drain is placed.²⁹ Surgical management of chyle leakage is by thoracic duct ligation. Conservative treatment has got the risk of loss of fluid approx up to 2.5 L daily protein, fats and lymphocytes, and immunological abnormalities.²⁸

There are always chances of it becoming empyema with secondary infection. In addition, there are possibilities of late complication like loculated chyloma or fibrothorax. In chylothorax secondary to malignancy, tetracycline or talc pleurodesis may be tried or even pleurectomy may be done.²⁹⁻³³ In our case cardiac tamponade occurred which led to haemodynamic instability, pericardial catheter helped to drain chyle from pericardium.

References

1. Sassoon CS, Light RW. Chylothorax and pseudochylothorax. *Clin Chest Med* 1985;6:163-71.
2. Seriff NS, Cohen ML, Samuel P, Schulster PL. Chylothorax: diagnosis by lipoprotein electrophoresis of serum and pleural fluid. *Thorax* 1977;32:98-100.
3. Goorwitch J. Traumatic chylothorax and thoracic duct ligation. *J Thorac Surg* 1955;29:467-79.
4. Cevese PG, Vecchione R, D'Amico DF. Postoperative chylothorax: six cases in 2,500 operations, with a survey of the world literature. *J Thorac Cardiovasc Surg* 1975;69:966-70.
5. Lam KH, Lim STK, Wong J, Ong GB. Chylothorax following resection of the oesophagus. *Br J Surg* 1979;66:105-9.
6. Wiener ES, Owens L, Salzberg AM. Chylothorax after Bochdalek herniorrhaphy in a neonate. *J Thorac Cardiovasc Surg* 1973;65:200-6.
7. Higgins CB, Mulder DG. Chylothorax after surgery for congenital heart disease. *J Thorac Cardiovasc Surg* 1971;61:411-8.
8. Tandon RK. Chylothorax after repair of ventricular septal defect. *J Thorac Cardiovasc Surg* 1968;56:378-80.
9. Kaul TK, Bain WH, Turner MA, Taylor KM. Chylothorax: report of a case complicating ductus ligation through a median sternotomy, and review. *Thorax* 1976;31:610-6.
10. Joyce LD, Lindsay WG, Nicoloff DM. Chylothorax after median sternotomy for intrapericardial cardiac surgery. *J Thorac Cardiovasc Surg* 1976;71:476-80.
11. Weber DO, Mastro PD, Yarnoz MD. Chylothorax after myocardial revascularization with internal mammary graft. *Ann Thorac Surg* 1981;32:499-502.
12. Mulders AV, Lacquet LM, Mieghem WV, Deneffe G. Chylothorax complicating pneumonectomy. *Thorax* 1984;39:954-5.

13. Schmidt A. Chylothorax: review of 5 years' cases in the literature and report of a case. *Acta Chir Scand* 1959;118:5-12.
14. Bower GC. Chylothorax: observations in 20 cases. *Dis Chest* 1964;46:464-8.
15. Roy PH, Carr DT, Payne WS. The problem of chylothorax. *Mayo Clinic Proc* 1967;42:457-67.
16. Bessone LN, Ferguson TB, Burford TH. Chylothorax. *Ann Thorac Surg* 1971;12:527-50.
17. Macfarlane JR, Holman CW. Chylothorax. *Am Rev Respir Dis* 1972;105:287-91.
18. Restoy EG, Cueto FB, Arenas EE, Duch AA. Spontaneous bilateral chylothorax: uniform features of a rare condition. *Eur Respir J* 1988;1:872-3.
19. Flaherty S, Ellison R. Bilateral chylothorax following thymectomy: resolution following unilateral drainage. *Mil Med* 1994;159:627-8.
20. Hillerdal G. Chylothorax and pseudochylothorax. *Eur Respir J* 1997;10:1157-62.
21. Jeske W. Chylothorax in infancy. *Thorax* 1968;23:214-5.
22. Carrington CB, Cugell DW, Gaensler EA. Lymphangiomyomatosis: physiologic-pathologic radiologic correlations. *Am Rev Respir Dis* 1977;116:977-95.
23. Klempser RG, Berry JF. The diagnosis and surgical management of chylothorax with the aid of lipophilic dyes. *Dis Chest* 1954;25:409-26.
24. Hashim SA, Roholt HB, Babayan VK, Itallie TBV. Treatment of chyluria and chylothorax with medium chain triglyceride. *N Engl J Med* 1964;270:756-61.
25. Gershanik JJ, Jonsson HT, Riopel DA, Packer RM. Chylothorax: a review of 18 cases dietary management of neonatal chylothorax. *Pediatrics* 1974;53:400-3.
26. Craenen JM, Williams TE, Kilman JW. Simplified management of chylothorax in neonates and infants. *Ann Thorac Surg* 1977;24:275-7.
27. Maloney JV, Spencer FC. The nonoperative treatment of traumatic chylothorax. *Surgery (St Louis)* 1956;40:121-8.
28. Machleder HI, Paulus H. Clinical and immunological alterations observed in patients undergoing long-term thoracic duct drainage. *Surgery* 1978;84:157-65.
29. Ross JK. A review of the surgery of the thoracic duct. *Thorax* 1961;16:12-21.
30. Bruneau R, Rubin P. The management of pleural effusions and chylothorax in lymphoma. *Radiology* 1965;85:1085-92.
31. Gingell JC. Treatment of chylothorax by producing pleurodesis using iodized talc. *Thorax* 1965;20:261-9.
32. Selle JG, Snyder WH, Schreiber JT. Chylothorax: indications for surgery. *Ann Surg* 1973;177:245-9.
33. Adler RH, Levinsky L. Persistent chylothorax: treatment by talc pleurodesis. *J Thorac Cardiovasc Surg* 1978;76:859-64.