

Transient spontaneous chylothorax presented with neck pain and swelling in patient with latent TB Case report

Hussam Almasri¹, Ans Alamami¹, Riyadh Hammamy¹, and Mohamed Shariff¹

¹Hamad Medical Corporation

October 19, 2020

Abstract

chylothorax is a relatively uncommon condition. It is characterized by the presence of chylous rich fluid within the pleural space. Spontaneous chylothorax usually refers to non-traumatic causes most common malignancy and infections. we present a case of a spontaneous chylothorax with evidence of resolution of pleural effusion without detectable cause.

Introduction:

Spontaneous chylothorax is a rare medical condition in which accumulation of chyle-containing lymphatic fluid in the pleural cavity happens with no history of trauma. In general, chylothorax is associated with significant mortality and morbidity up to 10%.

Chylothorax is defined by the presence of high triglycerides and low cholesterol level in lymphatic exudative pleural effusion, most of the times the fluid appears milky on aspiration.¹

The aetiology of chylothorax varies from neoplasm to infections like tuberculosis and many other causes.⁹

Transient spontaneous chylothorax was reported in few cases and was suggested to be due to minimal injury to the thoracic duct like sudden head movement and minimal exercise.¹⁰

Here we present a case of young lady who suffered from neck pain and found to have transient chylothorax without history of trauma.

Our aim is to highlight the importance of this diagnosis and to avoid extensive testing in mild cases.

Case Presentation:

A 35 years old lady, not known to have chronic medical illness. Presented to the hospital with chief complaint of left sided neck swelling and pain of 2 days duration. She didn't have a history of fever, cough, weight loss, haemoptysis, chest pain or shortness of breath. The patient reported no previous history of tuberculosis in the family or any sick contacts, and this was the first time to have such symptoms. The patient is a reformed smoker and drinks socially. There was no past history of trauma or any vigorous exercise.

Upon arrival to the emergency room, she was afebrile, blood pressure 104/64 mmgh, respiratory rate 18/minute, pulse rate 83/minute.

Neck examination revealed left supraclavicular swelling, tender but soft with no palpable lymph nodes, examination of the chest revealed stony dullness and reduced breath sounds in the basal left zone.

Ultrasonography revealed ill-defined predominantly hyperechoic mixed echogenic area in the left supraclavicular region, and left sided pleural effusion.

CT neck and chest revealed diffuse fat stranding and small lymph nodes noticed in mediastinum giving picture of mediastinitis/ Inflammatory process involving the left posterior neck muscle and in left pectoralis muscle with diffused smudged fat plane.

No collection noted in the neck. Mild left pleural effusion suggestive of chylothorax, figure 1, figure 2.

Laboratory investigations including complete blood count, comprehensive metabolic profile and C-reactive protein, lipase, thyroid function tests were all within normal limits.

Serum triglyceride 1.9 mmol/L, normal limit 1.7 to 5.6 mmol/L. Serum cholesterol 4.1 mmol/L, normal limit 5.2 to 6.2 mmol/L.

Ultrasound guided diagnostic aspiration of the pleural fluid showed milky alkalotic exudative fluid with predominant lymphocytes, triglycerides level of 2.39 mmol/l, cholesterol level of 3.4 mmol/l, negative gram stain and bacterial culture, negative acid-fast bacilli smear, culture and TB-PCR, and also negative cytology. QuantiFERON was positive.

The patient's symptoms started to improve during hospital stay with symptomatic treatment.

Few days later, the patient underwent repeated CT scan which showed resolution of most of the pleural effusion with normal abdomen CT findings, figure 3.

Follow up chest X-ray after one month was unremarkable and the patient was free of symptoms

Discussion:

Chylomicrons arise from the combination of long chain triglycerides with cholesterol esters and phospholipids. These molecules are resistant to be broken down by intestinal lipase so they pass to the lymphatic system of the small intestines then via thoracic duct to systemic circulation.¹

The thoracic duct length varies from 38-45 cm and ends typically at the junction between internal jugular and left subclavian veins carrying lymph from lower limbs and chyle from intestines.¹⁰

Chylothorax results from the leak of chyle from thoracic duct to the pleural cavity. This can happen after rupture, disruption or obstruction of the thoracic duct.¹

The aetiology of chylothorax can be categorized into traumatic, non-traumatic (spontaneous) and idiopathic. Chylothorax usually present with acute symptoms including dyspnoea, fatigue and less commonly chest pain and fever. Chyle is a non-irritating fluid to the pleura and this can explain the infrequency of chest pain upon presentation.

The diagnosis of chylothorax is mainly based on aspirated fluid analysis, the milky appearance is not exclusive to chylothorax, other conditions like empyema and cholesterol effusion can cause milky pleural effusion and the differentiation between these conditions is vital for management.^{8,10}

Chylothorax is defined by the presence of triglycerides more than 110 mg/dl in the fluid and cholesterol less than serum. Usually the fluid is alkalotic, lymphocytic and exudative,^{8,10} but it was reported to be transudative rarely¹⁵

Previously, non-traumatic chylothorax especially from neoplasms was the most common in adults but recent reports indicated that traumatic mainly post-operative cases are more common, this can be due to increased number of chest procedures and surgery or only increased recognition.^{1,4}

Traumatic chylothorax was described in various types of surgery including chest, neck, cardiac, gastric and oesophageal. It is considered to be iatrogenic with favourable outcomes in most cases. Oesophageal surgery was considered the highest risk surgery to develop chylothorax. In addition to surgery, blunt trauma and penetrating injuries were also associated with chylothorax due to direct or indirect thoracic duct damage.^{1,4}

Most cases of non-traumatic or spontaneous chylothorax comes from malignancy with lymphoma to be most common followed by bronchogenic carcinoma and other tumours, kaposi sarcoma was also reported.⁴

Other than neoplasms, many causes were identified to result in chylothorax including tuberculosis, filariasis, sarcoidosis, congestive heart failure, yellow nail syndrome, lymphangioleiomyomatosis, lymphatic malformation and radiation therapy.^{3,4,12}

Congenital chylothorax was described in new-borns to be the most common cause. Trisomy 21 or Turner syndrome appear to be associated risk factor.

Spontaneous chylothorax with no apparent cause was reported in few cases, minimal physical activity or sudden head movement especially neck hyperextension was thought to be the precipitating factor along with recurrent vomiting, hiccups and cough.^{9,11} It was suggested that there should be a weak point from pre-existing disease to cause thoracic duct rupture with minimal exercise, one of the cases had previous TB infection and like our case⁹, the presence of positive QuantiFERON raise the possibility of latent TB which could have played a role in causing disruption of the thoracic duct. There was no evidence of active TB in our patient and the spontaneous resolution of pleural fluid can support that.

Management of Chylothorax can be either conservative or surgical. Conservative treatment includes the use of a low-fat diet supplemented with medium chain triglycerides (MCT), other interventions are available based on the case including chest tube drainage, pleurodesis, thoracic duct ligation or embolization.^{5,7}

But it's worthwhile to know that most of the cases of spontaneous chylothorax are self-limiting and can be managed conservatively with rest, good hydration and low-fat diet.

Conclusions: spontaneous chylothorax can be the result of variety of conditions. It should be always in mind that some cases are transient and no underlying disorder can be identified. We recommend higher threshold for invasive investigations when no alarm signs are detected.

Patient Perspective: 'it was the first time having these symptoms. I was relieved that the illness is benign and will not need more procedures.'

Author contribution:

Hussam Almasri MD: manuscript writing, literature review, correspondence

Riyadh Hammamy: literature review, manuscript writing

Ans Alamami: literature review

Khalid Shariff: clinical follow up

All authors declare no conflict of interest

Ethical Approval: Medical Research Center, Hamad Medical Corporation, MRC-04-20-628

Patient consent has been obtained for publication of case details.

Funding information: funded by Qatar National Library

Acknowledgment: Dr Hatem Abdulkareem Abdulmajeed, Hamad Medical Corporation, Radiology Department for helping in image identification and extract.

References

1. Rudrappa M, Paul M. Chylothorax. [Updated 2019 Dec 4]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2020 Jan-. Available from: <https://www.ncbi.nlm.nih.gov/books/NBK459206/>
2. Kutlu O, Demirbas S, Sakin A. Chylothorax due to tuberculosis lymphadenitis. *North Clin Istanbul*. 2017;3(3):225-228. doi: 10.14744/nci.2015.52714
3. Bielsa S, Pardina M, Porcel JM. Chylothorax due to enlarged tuberculous lymph nodes. *BMJ Case Rep*. 2014;2014. doi: 10.1136/bcr-2014-204582

4. Doerr CH, Allen MS, Nichols FC, Ryu JH. Etiology of chylothorax in 203 patients. *Mayo Clin Proc.* 2005;80(7):867-870. doi: 10.4065/80.7.867
5. Zabeck H, Muley T, Dienemann H, Hoffmann H. Management of Chylothorax in Adults: When is Surgery Indicated? *Thorac Cardiovasc Surg.* 2011;59(04):243-246. doi: 10.1055/s-0030-1250374
6. Mortman KD. Mediastinal Thoracic Duct Cyst. *The Annals of Thoracic Surgery.* 2009;88(6):2006-2008. doi: 10.1016/j.athoracsur.2009.04.138
7. Nadolski G. Nontraumatic Chylothorax: Diagnostic Algorithm and Treatment Options. *Techniques in Vascular & Interventional Radiology.* 2016;19(4):286-290. doi: 10.1053/j.tvir.2016.10.008
8. Agrawal V, Doelken P, Sahn SA. Pleural Fluid Analysis in Chylous Pleural Effusion. *CHEST.* 2008;133(6):1436-1441. doi: 10.1378/chest.07-2232
9. Herzog KA, Branscom JJ. Spontaneous Chylothorax. *CHEST.* 1974;65(3):346-347. doi: 10.1378/chest.65.3.346
10. Schwartz J. Spontaneous Chylothorax. *JAMA.* 1960;174(16):2033-2035. doi: 10.1001/jama.1960.03030160019005
11. Torrejais JCM, Rau CB, de Barros JA, Torrejais MM. Spontaneous chylothorax associated with light physical activity. *J Bras Pneumol.* 2006;32(6):599-602. doi: 10.1590/s1806-37132006000600021
12. Dagenais F, Ferraro P, Duranceau A. Spontaneous chylothorax associated with primary lymphedema and a lymphangioma malformation. *The Annals of Thoracic Surgery.* 1999;67(5):1480-1482. doi: 10.1016/S0003-4975(99)00219-2
13. Hamad WB, Amar JB, Dhahri B, et al. Spontaneous chylothorax revealing a mediastinal and abdominal lymph node tuberculosis. *European Respiratory Journal.* 2014;44(Suppl 58). Accessed June 25, 2020.<https://erj.ersjournals.com/content/44/Suppl.58/P2648>
14. Valentine VG, Raffin TA. The Management of Chylothorax. *CHEST.* 1992;102(2):586-591. doi: 10.1378/chest.102.2.586
15. Diaz-Guzman E, Culver DA, Stoller JK. Transudative Chylothorax: Report of Two Cases and Review of the Literature. *Lung.* 2005;183(3):169-175. doi: 10.1007/s00408-004-2531-2

Figure legends:

Figure 1. CT scan of the chest showing left-sided pleural effusion with high attenuation suggestive of chylothorax

Figure 2. CT scan showing left neck subcutaneous oedema, the track of thoracic duct.

Figure 3. Follow up CT scan showed resolution of the left sided pleural effusion.



