

## A Rare Case of Sudden-Onset Respiratory Distress in an Infant

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### ABSTRACT

Chylothorax is an unusual cause of pleural effusion after the neonatal period in children without a previous history of cardiothoracic surgery. Determination of the causes of this condition can be often challenging. Herein, we present a case of a 6-month-old male infant, presenting with sudden severe breathlessness, which was later diagnosed as spontaneous left-sided chylothorax.

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### Introduction

Spontaneous chylothorax in children, especially after the neonatal period, is a rare condition. Herein, we introduce a case of a 6-month-old male infant, presenting with acute respiratory distress syndrome. Pleural fluid tap revealed a chylous effusion and the infant was treated by intercostal drainage, octreotide infusion and a medium-chained triglyceride (MCT)-based diet, leading to gradual resolution of chylothorax.

### Case report

A 6-month-old male infant was referred to the hospital with acute breathlessness over the past 7 days. He was afebrile with no history of cough, coryza, trauma to the chest or spine, weight loss, vomiting or lethargy. He was a full-term infant with no perinatal complications. No dysmorphic features were present and there was no previous history of contact with tuberculosis.

Based on the examinations, the patient's temperature, heart rate, respiratory rate and blood pressure were 36.9°C, 150 bpm, 80 bpm, and 92/62 mmHg, respectively. Severe respiratory distress, with subcostal and intercostal retractions,

was noted. The chest was dull to percussion, with poor air entry to the left hemithorax. The rest of the physical examination was unremarkable.

Chest X-ray was carried out, which showed opacification of left hemithorax with mediastinal shifting to the right. Needle aspiration was also performed, which revealed milky white odourless fluids. Pleural fluid analysis showed a cell count of 1300 cells/ $\mu$ l with 90% lymphocytes and triglyceride level of 1781 mg/100 ml, suggestive of chyle; thus, spontaneous chylothorax was diagnosed.

Under local anaesthesia, an intercostal catheter was placed for drainage (Figure 1). Chest computed tomography excluded the presence of mediastinal mass and confirmed the presence of a massive pleural effusion (Figure 2). Tests were negative for tumour markers including  $\alpha$ -fetoprotein and  $\beta$ -human chorionic gonadotropin; in addition, mantoux test result was negative (diameter: 1 mm). Radionuclide lymphoscintigraphy was carried out after 20 days of admission, which confirmed the normality of lymphatic system.

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**Figure 1.** Chest Xray showing left side pleural effusion with intercostals drain in situ

The patient was kept on oxygen support, intravenous (IV) antibiotics and total parenteral nutrition. Pleural fluid culture showed no growth and IV antibiotics were withdrawn. We started the patient on octreotide infusion 5µg/kg/hr. Pleural fluid loss decreased from about 11ml/kg/hr to 2 ml/kg/hr within 6 days. As the patient's condition improved, he was started on an MCT-based diet.

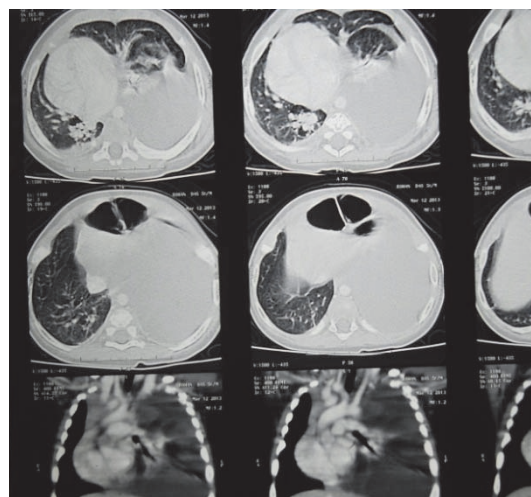
The intercostal drain was removed after 14 days of admission and repeated chest X-ray showed complete lung expansion, with no fluid collection. The infant was switched over to a normal diet and discharged on the 28<sup>th</sup> day of admission. Chest X-ray of the infant showed a centrally located heart and the echocardiography confirmed our diagnosis of mesocardia; however, there were no other associated cardiac or systemic anomalies.

## Discussion

Chylothorax is a rare cause of pleural effusion in children. Cases of spontaneous chylothorax after the neonatal period have been rarely reported in medical literature.

Damage to the thoracic duct causes the accumulation of chyle in the pleural space. Chyle refers to lymphatic fluid enriched with fat (chylomicrons), which is absorbed by intestinal cells and circulated via the thoracic duct (1, 2). This condition is common among newborns as a congenital condition, secondary to birth trauma.

In childhood, chylothorax generally occurs after cardiac surgery. Other causes include neck surgery, scoliosis surgery, congenital malformations of pulmonary or thoracic lymphatic systems and dysmorphic syndromes (Turner's, Noonan's and Down syndromes) (2, 3). Some of the other known causes include blunt trauma to the chest, hyperextension of thoracic spine, forceful vomiting/coughing, subclavian



**Figure 2.** CT Scan showing massive left side pleural effusion with mediastinal shift

vein thrombosis, superior vena caval thrombosis and thoracic duct compression or infiltration due to malignancies such as lymphoma, sarcoma, neuroblastoma, teratoma, tuberculosis and sarcoidosis (4). In our case, none of the known causes of chylothorax could be identified, despite extensive examinations.

Knowledge of the anatomy of thoracic lymphatic system is helpful in determining the site of thoracic duct disruption, associated with right-sided, left-sided or bilateral chylothorax. Rupture of the thoracic duct between the diaphragm and 5<sup>th</sup> thoracic vertebra (T5) usually produces a right-sided chylothorax; on the other hand, rupture of the thoracic duct above T5 leads to a left-sided chylothorax (5, 6). When bilateral chylothorax occurs, damage of the duct is usually part of a more diffuse lymphatic condition or located where the duct passes the mid-line at the level of T5 (7). In our case, there was a left-sided chylothorax, unlike other previously reported cases of spontaneous right-sided chylothorax (6).

In the present case, lymphoscintigraphy indicated no leakage, which is probably related to the completion of healing process by the time the scan was performed; the scan could not be carried out earlier due to financial constraints.

The basic principle of chylothorax management (irrespective of the cause) is to reduce chyle flow in the thoracic duct, while waiting for spontaneous healing. This is usually managed by a low-fat, MCT-based diet or occasionally, enteric rest with total parenteral nutrition. Spontaneous healing can take weeks to complete. MCT contain fatty acids which are 8-12 carbons in length and are absorbed directly in the portal venous system, bypassing the lymphatic drainage (8).

Use of octreotide, a somatostatin analogue, has been recently advocated for children with chylothorax, who do not respond to conventional

therapy (9, 10). Although its mechanism of action in treating chylothorax is unclear, reduced splanchnic vascular tone, which eventually leads to decreased flow of chyle through the thoracic duct, might be a possible underlying mechanism (11, 12). Our case was also diagnosed with mesocardia on echocardiography. After extensive search on the internet by Google engine, we could not find any published studies reporting an association.

### Conflict of Interest

Authors declare no conflict of interest.

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