

# Successful surgical intervention in a case of spontaneous chylothorax in an infant unresponsive to conservative therapy

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**Abstract:** We report a case of an infant with spontaneous chylothorax due to the congenital malformation of a small lymph vessel of the chest wall. Conservative therapy with omitting long-chain fatty acids from the diet, fat-free nutrition, total parenteral nutrition and intravenous somatostatin did not result in the decrease of pleural effusion. Thoracic surgical intervention performing thoracic duct ligation and using fibrin sealants was applied after 10 days of unsuccessful conservative therapy, and resulted in the complete recovery of the patient. Our experience support the already existing observations, that in cases where the daily loss of chyle exceeds 100 ml per age years and/or lasts longer than 2 weeks, early surgical intervention is recommended.

**Keywords:** chylothorax, lymphatic malformation, thoracic duct, somatostatin

## Introduction

Spontaneous chylothorax in childhood is a rare condition with a high mortality rate and no established treatment options [1], resulting mainly from malignancies, subclavian vein thrombosis, thoracic surgery, trauma, and infectious diseases [2, 3]. It is rarely due to the congenital malformation of the thoracic lymphatic system [4]. Authors report a case of an infant with spontaneous chylothorax due to the congenital malformation of a small lymph vessel of the chest wall.

## Case report

A 4-month-old infant presenting faintness, respiratory distress and feeding problems was admitted. Chest X-ray and CT scan (*Fig. 1*) showed homogeneous opacity on the complete left side of the chest with a remarkable midline shift, suggesting the presence of pleural fluid. Bronchoscopy revealed no intrabronchial lesions. Diagnostic thoracocentesis was performed, and 300 ml milky pleural fluid was drained. The laboratory analysis of the pleural fluid revealed high triglyceride (3.9 mmol/L) and protein (25 g/L) content, as well as high leukocyte (24.91 G/L) count, which was chyle according to the Light's criteria [5]; and to the guidelines of Buttiker et al.

[4] (*Fig. 2*). To avoid respiratory failure, continuous pleural drainage was introduced.

On days 1–3 partial parenteral nutrition and long-chain fatty acid free hydrolyzed formula was used. Latter is known to preferably change the constitution of chyle and prevent malnutrition, [6] simultaneously decreasing chyle leakage, although latest reports suggest that it is of no use in the clinical management of non-traumatic chylothorax [7]. As no decrease in the chyle effusion was achieved, enteral nutrition was changed to fat-free diet complemented with parenteral nutrition for another 2 days. On day 5 post admission, enteral nutrition was totally omitted, calorization was achieved via total parenteral nutrition, and intravenous somatostatin was introduced in a dose of 20 µg/kg/day [8]. After 5 days of such regime the pleural fluid became transparent, but its quantity did not decrease – a daily amount of 300–580 ml was drained (54.54 ml/kg body weight – 105.45 ml/kg body weight). By day 9, the nutritional and cardiopulmonary status of the patient worsened, and threatened with fatal outcome. Considering that the persistent loss of such amount of lymph containing sugars, fats and immunologically active cells may lead to an irreversible energetic and immunological decline, surgical termination of lymph leakage was indicated, according to Buttiker et al. [4].



Fig. 1. ■ Chest CT scan of the patient. Pleural effusion covers the left side of the chest causing midline shift

On day 5 of total parenteral nutrition and somatostatin treatment (day 10 post admission), 3-port video-assisted thoracoscopy was performed to find and eliminate the leakage site. General anesthesia using double-lumen endotracheal tube utilizing single-lung selective ventilation was administered. Leakage site in the posterior sinus was

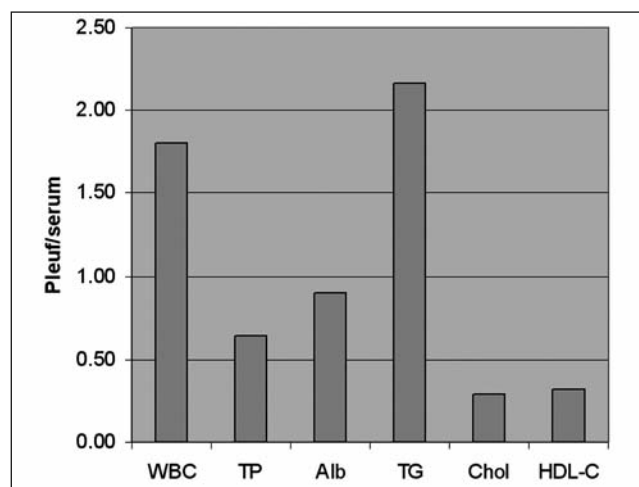


Fig. 2. ■ Pleural/serum concentration ratios of constituents of the pleural fluid seen in chylothorax. WBC: white blood cells, TP: total protein, Alb: albumin, TG: triglyceride, Chol: total cholesterol, HDL-C: high density lipoprotein bound cholesterol. Pleural/serum triglyceride ratio is 2.17 suggesting chylous origin

detected using intravenous soybean oil (Intralipid, Fresenius Kabi, Lajos u. 48–66, H-1036 Budapest, Hungary) where an aberrant lymph vessel of the chest wall was identified at the level of the 10<sup>th</sup> rib. The aberrant lymph vessel and the thoracic duct was ligated, resulting in the complete cessation of lymph leakage. Prior to the closure of the chest, fibrin sealant coating was applied over the affected chest wall area.

Continuous pleural suction was maintained until the 7<sup>th</sup> postoperative day, fat-containing enteral nutrition was administered on the postoperative 3<sup>rd</sup> day. No further chylous leakage was seen, however, on the postoperative 3<sup>rd</sup> day, adult respiratory distress syndrome developed. Latter was successfully treated with diuretics, steroid and mechanical ventilation using positive end-expiratory pressure. Patient recovered fully.

## Discussion

Spontaneous chylothorax resulting from congenital lymph vessel anomalies is a rare condition in the pediatric age group, requiring a multidisciplinary therapeutic approach. Continuous chyle leakage with loss of fats and lymphocytes threatens with a potentially fatal malnutrition and immunosuppression [9]. Our experiences support some already existing observations, that where the daily loss of chyle exceeds 100 ml per age years and/or

lasts longer than 2 weeks, conservative therapy is of limited indications, early surgical intervention is recommended [10, 11].

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