Karyotyping remains the gold standard for detecting chromosomal aberrations in cases with congenital anomalies. A meaningful correlation between the deletion and the clinical phenotype is not possible until further use of high-resolution investigations like CGH array to fully characterize the case, which was not possible due to financial constraints.

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REFERENCES


Thoracoscopic Ligation of Thoracic Duct for Spontaneous Chylothorax

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Spontaneous chylothorax, without a predisposing factor is an uncommon cause of pleural effusion beyond the neonatal period. We present a case of left sided spontaneous chylothorax in a 20-month-old boy. We report successful management of this difficult problem with thoracoscopic ligation of thoracic duct after a failed trial with conservative management.

Keywords: Chylothorax, Thoracoscopic, Thoracic duct, VATS.

chylothorax has various causes, including malignancy, trauma (including surgery), and miscellaneous disorders (such as deep vein thrombosis, sarcoidosis, and congestive heart failure), and can also be idiopathic [1,2]. Undetected malformations of the lymphatic trunks are implicated as a cause of spontaneous chyle accumulation. Management of spontaneous chyle accumulation in a child is a challenging task. We present a child with left sided spontaneous chylothorax who was managed with thoracoscopic ligation of thoracic duct on right side.

CASE REPORT

A 20-month-old boy presented with fever and breathing difficulty for one week. There was no history of trauma or operative intervention in the child. The mother gave no history of excessive cough or vomiting. The child was otherwise healthy, with no significant past medical history. There was no history of recent trauma or history suggestive of cardiopulmonary disease. The child’s immunization was up to date.

On examination, the child weighed 12 kg, was febrile,
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Pulse rate was 130/minute, blood pressure was 90/64 mm Hg and respiratory rate was 56/minute. His blood biochemistry and hematological parameters (total leucocyte count was 9600 with 80% neutrophils, 12% lymphocytes, 7% monocytes and 1% eosinophils) were all within normal limits. The CRP level was 1.8 mg/L and ESR was 4 mm. Chest examination revealed absent breath sound on left side. Chest X-ray revealed an opaque left hemithorax. Computed tomography of chest showed massive left sided pleural effusion with mediastinum shifted to right side. There was no mass or any other abnormality. Left side intercostal drain (24 Fr) was inserted and 640 mL milky fluid was drained from the chest. Fluid analysis revealed total cell count of 10000 per cubic mm with lymphocytic predominance, and high triglyceride (150 mg/dL). He was started on intravenous antibiotics and octreotide infusion at 240 μg/day in 12 cc NS at the rate of 0.5 mL/h for 7 days. He became afebrile with the treatment but chest tube continued to drain 600-700 mL of milky fluid every day. Lymphoscintigraphy performed after 5 days of conservative approach showed a large leak of dye in left pleural space; however, the site of leak could not be identified. The chest tube output and character remained unchanged despite starting the child on fat restricted diet. Subsequently, oral feeds were stopped and total parenteral nutrition (TPN) was started; octreotide was continued for further two weeks. The chest tube output reduced to 400 mL/day of rice water color fluid in the first few days and remained unchanged thereafter. A surgical consultation was sought when conservative treatment failed even after 3 weeks. At this stage, thoracoscopic ligation of thoracic duct on right side was offered.

The procedure was performed under general anesthesia. Selective deflation of right lower lobe was achieved using Fogarty balloon tipped catheter inserted through the ETT and advanced to the right lower lobe bronchus under fiber optic bronchoscopy guidance. The child was placed supine with 30 degrees right up position. Three 5-mm ports were used: the port positions included the mid-axillary line 5th intercostal space, mid clavicular line 7th intercostal space, and posterior axillary line 7th intercostal space. The procedure was begun with incision of mediastinal pleura between azygous vein and thoracic aorta in the lower chest (just above the diaphragm). The thoracic duct was identified as a thin walled tubular structure lying between the azygous vein and the aorta. Butter milk was given to the child through nasogastric (Ryle’s) tube to make the duct prominent for visualization. Metallic clips were applied to occlude the duct. The milky output through the left chest tube stopped immediately after clipping of the duct and the closure done after insertion of 20 Fr chest tube into right chest. The child tolerated the procedure well, was extubated on the table and allowed orally from the next day. The right chest tube drained 100 mL of serosanguineous fluid on the first post-operative day. It was removed on the second post-operative day when the drainage was 40 mL. Despite normal food intake, the left chest tube output started reducing with each passing day, stopping completely on day 14 and the tube was removed on day 16. The child was discharged the next day. He has been on normal diet and is doing well at 6-months follow up.

DISCUSSION

Chylothorax needs prompt treatment. Drainage of the pleural cavity by chest-tube relieves the patient of breathing difficulty. Thereafter, conservative treatment should be started, consisting of restriction of dietary fat to medium-chain triglyceride and fluid and electrolyte replacement. If there is no improvement, all oral intakes should be stopped and total parenteral nutrition should be implemented [2-4]. Although, there is no uniform agreement in medical literature, several authors have reported successful treatment of cases of congenital and postoperative chylothorax in children with octreotide infusion [4,5]. TPN has its own attendant complication and it is a challenging task to keep a 20-month-old child nil orally for a long time.

If loss of chyle persists, surgical treatment should be considered. Pleurodesis with talc or povidone-iodine, fluoroscopically guided embolization of the thoracic duct, pleuro-peritoneal shunt and pleurectomy have been tried with variable success. Surgical ligation of the thoracic duct represents the most definitive treatment of chylothorax. There are a very few reports of use of thoracoscopic thoracic duct ligation for spontaneous chylothorax in a child in the English literature. We could find only two reports describing the successful thoracoscopic ligation in such circumstances [6,7]. Martinez, et al. [7] subsequently published an erratum stating that there case report was not idiopathic but probably secondary to trauma [7]. In its course through the left chest, the thoracic duct is quite inaccessible. Hence it is accessed and ligated in the right lower chest, which takes care of left side leaks also. Some of these patients, however, do drain the chylous fluid, for few days, from the thoracic duct segment distal to the site of ligation, which closes in a few days. Traditionally, thoracic duct ligation in the right chest has been done by thoracotomy. Now, with the advancement of minimal access techniques, the same is possible through thoracoscopic approach, providing the benefits of minimal access surgery. Ample evidence in the literature
has proved the effectiveness of thoracoscopic ligation of thoracic duct in the management of chylothorax [8,9]. After thoracoscopic thoracic duct ligation, the output in our patient dropped to half of the usual output in the immediate postoperative period and decreased slowly over next few days to stop completely in 14 days. This was either due to leakage from the segment distal to the site of ligation (which healed slowly) or due to an accessory duct leaking into left pleural cavity.

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