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Congenital Chylothorax-A case report

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Abstract
Congenital chylothorax is an extremely rare condition. It is a rare cause of respiratory distress in the neonates, defined as a collection of chyle in the pleural space. Causes of chylothorax in neonates are congenital lymphatic malformations, associated syndrome or birth trauma. Here we describe a neonate who presented with severe respiratory distress require intubation and ventilator support and on evaluation no cause of chylothorax found. Baby managed with intercostal drainage and octreotide, improved and asymptomatic in 8 months of follow-up.

Keywords: chylothorax, neonate, octreotide

Introduction
Chylothorax is characterized by the accumulation of chylous fluid in the pleural space. It is an uncommon but is the most common cause of pleural effusion in the foetus and neonates. It is classically a disorder of infants at or near term [1, 2]. Males are affected twice as frequently as females and 60% of cases involve the right side of the chest. Congenital chylothorax usually occurs spontaneously due to lymphatic malformations or is associated with birth trauma to the thoracic duct, and it presents a clinical challenge; produces respiratory compromise, nutritional failure or immunologic depletion contributing to sepsis [1, 3]

Case Report
A 4 day old newborn presented to our department with refusal to feed and severe respiratory distress. The baby was full term, first born girl child delivered normal vaginally with a birth weight of 3.1 kg. There was no history of birth trauma. The baby was asymptomatic until day thirds of life with normal feed. The baby developed refusal to feed from the 4th day, thereafter she developed progressive respiratory distress. On examination, the baby was in severe respiratory distress with decrease air entry on the right side of the chest, no others any associated congenital anomalies were found. Baby wasintubated and kept on ventilator due to severe respiratory distress. Chest X-ray revealed right pleural effusion with mediastinal shift toward left. Contrast Enhanced Computed Tomography (CECT) thorax showed right pleural collection found with no contrast leak in the plural cavity, there were no signs of pleural inflammation, no any lymphatic malformation was found. Pleural tap was done which revealed milky white aspirate. On biochemical analysis of pleural fluid suggest chyle with triglyceride level of 120 mg/dl, pleural fluid to serum cholesterol ratio was <1.0 and chylomicron found. Cytological examination revealed absolute cell count 1100 cells/L, lymphocytes 800/mm^3, and erythrocytes 150/mm^3. Patient managed with intercostal drainage with the output of 300 ml initially, followed by 200 ml daily for 5 days. In view of persistent drainage in the tube in spite of diet, contacting low fat and containing medium chain triglycerides patient managed with long-acting somatostatin analog (octreotide) in dose of two micro gram/kg/subcutaneously six hourly. Response to octreotide noted on the first day of the start of therapy with gradual decreasing in drain output. On the 7th day of octreotide therapy, drainage stops and intercostal tube was removed in view of satisfactory chest condition in radiograph.

Discussion
The incidence of congenital chylothorax reported as 1 in 2000 neonatal Intensive Care Unit (NICU) admissions [4]. In idiopathic congenital chylothorax, neonates have a weak thoracic duct or lymphatic anomalies. Therefore, any increase in venous pressure may cause a break of the congenitally weakened thoracic duct. The mechanism thought to be either traumatic, with rupture of the thoracic duct by hyperextension of the spinal column or secondary to increased systemic venous pressure during birth, especially in complicated deliveries. Etiology is unknown in the majority of neonatal chylothorax cases as in our case in spites of relevant investigations. CECT may help in finding the anatomical cause of chylothorax, as lymphatic malformations can be detected. In cases of traumatic chylothorax, significant contrast leak may found in the pleural cavity. The idiopathic congenital chylothorax is usually a transient condition that resolves by conservative treatment with the dietary elimination of the long-chain fatty acids or replacement of oral feeding with total parenteral nutrition. Mortality rates range from 20% to 60% depending on associated findings, gestational age, and the duration and severity of the chylothorax.[6] In the case of continuation of drainage despite total parenteral nutrition, advocated to perform surgery like ligation of the thoracic ductus, pleuroperitoneal shunt, pleurectomy, or pleurosis not required in our casex [7].
Octreotide, a somatostatin analog, prescribed as an anti-secretory agent. Somatostatin reduces the intestinal

absorption of fats and decreases gut motility. Its mechanism of action in treating chylothorax is not clear, but a possibility is reduction of splanchnic vascular tone, eventually leading to a decreased flow of chyle through the thoracic duct. During therapy with octreotide, patient should be monitor for hyperglycemia, hypothyroidism, diarrhea, renal impairment, necrotizing enterocolitis, and liver dysfunction [8]. For management purpose, first, we should diagnose and determine the etiology of chylothorax, put an intercostal drainage tube for symptomatic improvement. In case drainage >100 ml/year/day, proceeds to surgical intervention and if drainage is less <100 ml/year/day, medical treatment can be tried. In cases were chylothorax persist for more than 2–4 weeks in spite of medical treatments, shift to surgical interventions.

Conclusion

Congenital chylothorax is a very rare cause of respiratory distress in neonates. Diagnosis of this condition is based on clinical, radiological, and pleural fluid biochemical findings. Conservative therapy should be tried first before a surgical procedure might be considered. Octreotide is an emerging agent in conservative therapy.

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