Perinatal/Neonatal Case Presentation

Spontaneous Neonatal Chylothorax Treated with Octreotide in Turkey: A Case Report

Ercan Sivasli
Deniz Dogru
Ayse Tana Aslan
Murat Yurdakok
Gulsevin Tekinalp

Chylothorax, which is usually idiopathic, is the most common form of pleural effusion encountered in neonates. Herein we report a 2-week-old infant who had spontaneous chylothorax and was treated with octreotide. Neonatal chylothorax responded well to octreotide treatment, which appeared to shorten the duration of hospitalization. We conclude that octreotide should be considered in the treatment of neonatal chylothorax.
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In infants and children, chylothorax may occur as a complication of surgery or birth trauma, in association with pulmonary tumors and pulmonary lymphatic abnormalities or in association with various syndromes. However, most commonly, the etiology remains unknown and the chylothorax is considered ''idiopathic''. Up to 50% of all incidents of chylothorax are recognized in the first week of life, but idiopathic neonatal chylothorax may be recognized even up to several weeks of age.[1,2]

Although thoracostomy drainage is the first-line therapy in the treatment of chylothorax, octreotide, a long-acting somatostatin analog that may act on somatostatin receptors in the splanchnic area to reduce lymph fluid production, has been used in chylothorax in infants and older children.[3,4] Herein we report a neonate who had spontaneous chylothorax and was treated with octreotide. To our knowledge, this is the second report in the literature.

CASE REPORT

A male newborn was delivered at the 34th week of gestation by cesarean section. The mother was 27 years old. She had received fraxiparine and acetylsalicylic acid during pregnancy due to antiphospholipid syndrome. Her triple test (α-fetoprotein, unconjugated estriol, and human chorionic gonadotropin) result was high, and karyotype by amniocentesis was 46, XY. There was first-degree consanguinity between the parents. Her previous obstetric history included a 30-week-old stillbirth, and three spontaneous abortions.

The birth weight of the neonate was 2070 g. His length was 48 cm and head circumference 34 cm. Clinical examination revealed low-set auricles, micrognathia, high palate, short neck, bilateral flexion contractures of the interphalangeal joints, and limitation of hip adduction. At the first day of life, surfactant was given because of respiratory distress syndrome and the patient was maintained thereafter on mechanical ventilation for 4 days.

Chest X-ray revealed infiltration on both sides, and pneumonia was diagnosed. He was treated with antibiotics and physical therapy was applied. On day 15, chest radiograph showed left pleural effusion. A thoracostomy tube was placed on the left side, and a milky-appearing fluid was drained. The fluid had the following characteristics: protein 2.6 g/dl, lactate dehydrogenase 370 IU/l, glucose 132 mg/dl, triglyceride 1602 mg/dl, cholesterol 58 mg/dl, chylomicron 17.4%, β-lipoprotein 11.5%, pre-β-lipoprotein 59.7%, α-lipoprotein 11.4%, and a high leukocyte count.

The neonate was diagnosed as having chylothorax. Enteral feeding was discontinued, and total parenteral nutrition was started. The amount of pleural drainage (120 ml on day 1) decreased gradually, and resolved at the 5th day. Then, the chest tube was removed and enteral feeding was restarted with human milk. At 2 days after the chest extubation, the chest tube had to be reinserted because pleural effusion reappeared on the left side. There was 5 to 6 ml milky-appearing pleural drainage. Therefore, intravenous infusion of octreotide, 3.5 μg/kg/hour daily, was started. At 72 h after institution of octreotide, pleural drainage stopped and the chest tube was removed. Blood glucose level was measured four times a day during full-dose octreotide therapy, and two times a day during half-dose octreotide therapy. Glucose level was normal in all of these measurements. Octreotide infusion was halved on days 29 and 30 and discontinued on day 31. Full enteral feedings with human milk were continued during octreotide infusion, and pleural effusion did not recur. After 2 weeks, the patient was discharged.

DISCUSSION

Although our case had an atypical face, he was nonsyndromic, and his karyotype was 46, XY. There was no known risk factor for the

Division of Neonatology, Department of Pediatrics, School of Medicine, Hacettepe University, Sihhiye, Ankara, Turkey.
Division of Pediatric Pulmonology, Department of Pediatrics, School of Medicine, Hacettepe University, Sihhiye, Ankara, Turkey.

Address correspondence and reprint requests to Ercan Sivasli, Division of Neonatology, Department of Pediatrics, School of Medicine, Hacettepe University, Sihhiye, Ankara 06100, Turkey.

occurrence of chylothorax in our patient. He had had no thoracic surgery, nor placement of a central venous catheter to cause thrombosis in the superior vena cava.[2] Therefore, our case was considered to be a spontaneous neonatal chylothorax.

Etiology is unknown in the majority of neonatal chylothorax cases. Idiopathic congenital chylothorax is mostly associated with lymphangiomatosis,[5] congenital lymphangiectasia,[6] Down's syndrome,[7] and maternal polyhydramnios.[8] These neonates are born with a weak thoracic duct or lymphatic anomalies. Therefore, any increase in venous pressure (e.g. during delivery) would lead to a break of the congenitally weakened thoracic duct.[3] Spontaneous neonatal chylothorax is usually a transient condition that resolves by cessation of the lymphatic flow in the thorax. In the patients, dietary elimination of the long-chain fatty acids or replacement of oral feeding with total parenteral nutrition can minimize production of the lymph, and thereby decrease lymphatic flow. However, this method causes a prolongation in the duration of pleural drainage, mechanical ventilation, and total parenteral nutrition. Meanwhile, the method leads to loss of lymphocytes, proteins, coagulation factors, and antibodies as well as lymphatic fluid, and causes an increase in the occurrence of complications like hypoproteinemia, coagulopathy, lenphopenia, hypogammaglobulinemia, sepsis, and ventilator-related pulmonary injury. In the case of continuation of drainage despite 2 to 5 weeks of total parenteral nutrition, it is advocated to perform surgery-like ligation of the thoracic ductus, pleuroperitoneal shunt, pleurectomy, or pleurosis.[1,9,10]

Use of somatostatin to treat chylothorax was first reported in 1990 in an adult,[11] and in 2003 in a neonate.[1] Somatostatin is a peptide that acts as a neurohormone. It inhibits pituitary secretion of thyroid-stimulating hormone and growth hormone, vasoactive intestinal peptide, gastrin and motilin in the gastrointestinal tract, as well as pancreatic secretion of insulin, glucagons, and somatostatin. Octreotide is a synthetic somatostatin analog. Although the exact mechanism of action of octreotide is not understood, it may act on somatostatin receptors in the splanchnic circulation, and decrease lymph fluid production through a reduction in gastric, intestinal, and pancreatic secretions or by a decrease in hepatic venous pressure and splanchnic blood flow. Somatostatin has also been used in children to treat diarrhea, enteric fistula, pancreatitis, and overgrowth, and in neonates to treat hyperinsulinism.[1,4,12,13]

The chylothorax in our case resolved completely, and no adverse effect was encountered. Thus, we report the successful use of octreotide in neonatal chylothorax. Standard management of neonatal chylothorax usually entails prolonged hospitalization and frequently requires surgical intervention. Administration of octreotide in our case led to a more rapid resolution of pleural drainage, no recurrence, and early hospital discharge. Total parenteral nutrition was discontinued promptly, and early initiation of enteral feeding was achieved. The use of octreotide therapy in neonatal chylothorax merits further investigation.

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