Clinical Brief

Congenital Chylothorax – Successful Management with Chemical Pleurodesis

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ABSTRACT

Congenital Chylothorax is a rare entity which is characterized by abnormal accumulation of chyle in pleural cavity. Chylothorax presenting as non-immune hydrops is even rarer. We report a case of congenital bilateral chylothorax presenting as non immune hydrops and managed successfully with chemical pleurodesis. A term male baby presented at birth with bilateral pleural effusions and subcutaneous edema. It was initially managed with ventilation and intercostals drainage (ICD). After the initiation of feeds, re-accumulation of pleural fluid led to the diagnosis of congenital chylothorax. Management with ICD and octreotide was unsuccessful but responded to chemical pleurodesis with 4% povidine iodine done on 3 separate occasions. **[Indian J Pediatr 2010; 77 (3) : 332-334]** *E-mail: srinivas_murki2001@ yahoo.com*

Key words : *Congenital Chylothorax; Octreotide; Chemical pleurodesis*

Congenital Chylothorax, an abnormal accumulation of chyle in the pleural space, is a common cause of pleural effusion in the neonatal period. Bilateral chylothorax is rarely seen. Chylothorax presenting as non-immune hydrops is even rarer. Management involves supportive care, parenteral nutrition, octreotide infusion and if necessary chemical or surgical pleurodesis. We report a case of congenital bilateral chylothorax presenting as non immune hydrops and managed successfully with chemical pleurodesis.

REPORT OF CASE

A 3880gm, term female baby, was born to a fifth gravida mother by elective cesarean section in view of previous three cesarean sections and polyhydramnios. Four weeks prior to admission, mother's examination and antenatal scan was reported normal. At birth, the baby girl was limp and asphyxiated. It was resuscitated with tube and bag ventilation and shifted to the neonatal intensive care unit (NICU). It was cyanosed and had generalized subcutaneous edema. There was no dysmorphic facies. The baby was connected to the ventilator and Radiograph chest showed bilateral pleural effusion (Fig. 1). Intercostal tubes (ICD) were inserted and nearly 200 ml of straw colored pleural fluid was drained from both the sides. Pleural fluid analysis revealed protein of 2.5g/dl, glucose of 75mg/dl, cell count of 28,100/cumm with 5% lymphocytes and 93% polymorphs. Culture was sterile. Ultrasound abdomen revealed normal liver, spleen and no ascites. 2D Echocardiogram was normal. The baby gradually improved, ICD tubes were removed on the fifth day,

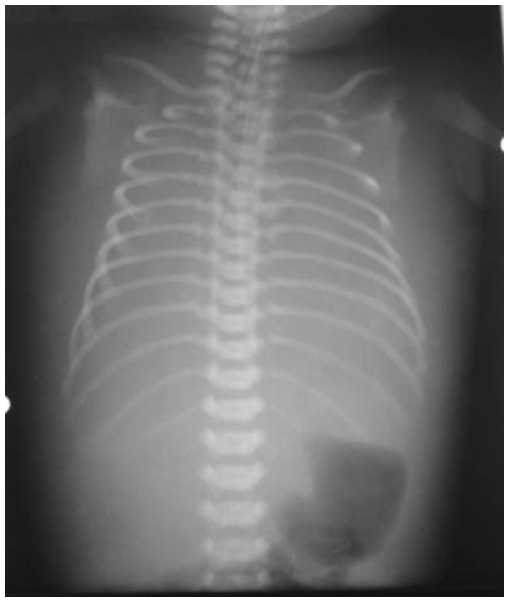

Fig. 1. Day one chest radiograph showing subcutaneous edema and bilateral pleural effusion.

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weaned off from the ventilator on sixth day, started on feeds on the same day and shifted to mother's side on the eighth day of life. On the ninth day, baby was again shifted back to the NICU in view of respiratory distress. Radiograph was suggestive of bilateral pleural effusion, left more than right. ICDs were re-inserted and analysis of fluid now revealed milky white fluid (Fig. 2) with triglycerides of 242mg/dl, cholesterol 22.4mg/dl, triglycerides to cholesterol ratio of 10.8, total counts of 34, 800/mm^3 with 4% polymorphs and 96% lymphocytes. Culture was sterile. A diagnosis of congenital chylothorax was made. Enteral feeds were discontinued and parenteral nutrition was instituted along with Octreotide infusion. Octreotide infusion was started at 1 µg/kg/hr and gradually increased to 10 µg/kg/hr. As the response was poor with octreotide, on the thirtieth and thirty-second day of life chemical pleurodesis was done with 4% povidine iodine. 4ml of 10% povidine mixed with 6ml of normal saline was injected through ICD tube into the pleural cavity and was retained for 5 hr. Octreotide infusion was tapered over next the five days. The baby improved with chemical pleurodesis and it was discharged home on oxygen at sixtieth day of life. A loculated collection of chyle was aspirated again on eightieth day of life. On follow up at 3months, the child is stable with no distress, good bilateral air entry and maintaining SpO2 > 95% without oxygen. Radiograph showed complete resolution of pleural effusions. Thyroid function is normal.

Fig. 2. Milky white fluid aspirated from the pleural cavity.

DISCUSSION

Congenital Chylothorax is a rare entity which is characterized by abnormal accumulation of chyle in pleural cavity. The index baby fulfilled all the criteria for diagnosis, pleural fluid protein concentration > 2 g/dl; Triglyceride concentration >100 mg/dl, number of cells/mm^3 > 100 with lymphocyte predominance and sterile culture. Milky appearance of the fluid (chylomicrons) is seen only in infants who are fed and at birth, it is straw colored as in this case.

A number of therapeutic interventions have been used to reduce chylothorax production and promote resolution. The primary interventions include avoiding dietary fat by using parenteral nutrition or medium chain triglyceride (MCT) rich formula feeds. We did not have access to MCT formulas and the baby was on parenteral nutrition for 25 days. The supportive care is very important in this condition as the disease process is prolonged and it involves multiple interventions. The ongoing losses further compromise the nutrition, thus protein replacement, adequate calories and electrolyte supplementation are important parts of management. Hypoalbuminemia needs to be corrected as there will be ongoing losses in chyle. These babies are at risk for sepsis as a large amount of prothrombin, fibrinogen, immunoglobulin and lymphocytes are lost in the chyle. Of late octreotide, a somatostatin analogue has become a good option for management. It is believed to reduce splanchnic blood flow which in turn decreases thoracic duct flow and triglycerides.[1] It can be administrated subcutaneously at 20–70 µg/kg/day in 3 divided doses or as an infusion at 1 to 10 µg/kg/hr. The reported duration of octreotide therapy ranges from 3 days to 29 days.[1] In the present case the response to octreotide was not satisfactory even at a dose of 10 µg/kg/hr.

Surgical treatment is advocated in resistant cases with excessive (> 10 ml / kg/day) or prolonged (> 3 – 4 weeks) drainage. The surgical options include chemical pleurodesis, pleurectomy, thoracotomy with thoracic duct ligation, intrapleural fibrin-glue or pleuro-peritoneal shunts.

Pleurodesis is one way to obtain pleural adhesion, thus obliterating chylous leaks. In a report by Brissaud O, *et al* chemical pleurodesis with povidine iodine was successful in three of the four neonates with congenital chylothorax.[2] They used 5ml to 10ml of either 4% betadine scrub or 10% betadine dermique. The duration of chest tube occlusion varied from 3 hr to 5 hr. In our case we used 10 ml of 4% betadine and chest tube was blocked for 5 hours. In another report by Mitanchez *et al* although chemical pleurodesis with betadine was successful, the infant developed renal failure.[3] In the present case the response to pleurodesis was good with no drainage except for loculated fluid which was treated with a third dose of betadine on the eightieth day of life. Thyroid function tests before and after chemical pleurodesis is recommended. Early diagnosis, aggressive initiation of non operative management and a number of surgical procedures have decreased the mortality rate from 50% before 1950's to 10–20% currently.

The present case is peculiar in its presentation as non immune hydrops, poor response to octreotide and parenteral nutrition but good response to chemical pleurodesis.

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