Neonatal Pleural Effusion Associated with Pulmonary Sequestration: A Case Report

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INTRODUCTION

Pleural effusion is an abnormal collection of fluid within pleural space usually resulting from excess fluid production and or decreased lymphatic absorption. Pleural effusion is rare during neonatal period with the estimated prevalence of 0.06%[1]. Neonatal pleural effusion may be due to antenatal congenital causes or postnatal acquired causes. Antenatal pleural effusion may be due to hydrops fetalis, congenital chylothorax, congenital heart disease (CHD), chromosomal anomalies, pulmonary anomalies or infection like congenital herpes simplex viral (HSV) and Paro virus infection. Postnatal acquired causes include extravasation of peripherally inserted central catheters, parapneumonic effusion or traumatic chylothorax.

As neonatal pleural effusion can cause significant respiratory distress, vigilant delivery room management is required. Identifying the underlying cause is important as it leads to specific therapeutic measures. Diagnostic chests tap for pleural fluid analysis is important for diagnosis and its consequent management.[2]Similarly, echocardiography, karyotyping and other investigations are also indicated for identifying subsequent etiology.

Here, we report a case of neonatal pleural effusion; which on evaluation; pulmonary sequestration-extra lobar type was found as the cause of effusion. Extra lobar pulmonary sequestration associated with pulmonary effusion is a very rare entity.[3] The purpose of this case report is to familiarize clinician regarding lung sequestration as the cause of neonatal pleural effusion and prevent subsequent morbidity and mortality.

CASE REPORT

A 22 years old G2P1L1 healthy female from Dang presented at 36+2 weeks of gestation (WOG) with leaking and labor pain. The antenatal scan was only done at 32 WOG which showed massive left sided pleural effusion. She was referred to Patan hospital for further evaluation and management. At Patan hospital ultrasonography (USG) scan was repeated which reconfirmed massive left sided pleural effusion without any other associated congenital anomalies. She delivered male baby weighing 2290gm via spontaneous vaginal delivery with Apgar score of 7/10 and 8/10. Baby developed grunting, sub costal retractions at 30 minutes of life. Baby was transferred to Neonatal Intensive Care Unit (NICU).

At NICU, baby was tachypenic with respiratory rate of 65/min and mild sub costal retractions with SPO_2 of 80% at room air; Downe's score was 4/10. There was no dysmorphism or any other congenital anomalies. Air entry was decreased on left lung. Cardiovascular examination was normal. There was no sign of heart failure. Abdomen was soft and there was no sign of fluid in the abdominal cavity. Child was started on non invasive positive pressure ventilation (NIPPV;RAM SIMV). Chest x-ray was done (Figure 1) which showed left sided pleural effusion. Chest tube was inserted (Figure 2) and around 200ml of pleural fluid was aspirated. Aspirated sample was also sent for analysis.

METHODS:

Hydrops fetalis, congenital chylothorax, congestive cardiac failure, fetal aneuploidy, structural fetal malformations like cystic adenomatoid malformations (CCAM), bronchopulmonary sequestration, congenital diaphgramatic hernia were sought as the possible differential diagnosis.

Evaluation to establish the cause began with pleural fluid analysis. Pleural fluid was straw coloured. Pleural fluid biochemistry showed protein 1.2gm/dl sugar 97mg/dl LDH 82U/L Triglyceride 13 mg/dl total cholesterol 50 mg/dl. Microscopic examination showed RBC: 10/mm^3 WBC 100/mm^{3}with polymorph of 30% and lymphocyte of 70%. Serum LDH and protein was 750U/L and 5gm/dl respectively, which showed pleural fluid to be non-chylous and transudative in nature. Pleural and blood culture were sterile. Echocardiography was done which showed small ASD; left to right shunt. No pericardial effusion and no signs of cardiac failure. Urine examination was normal with no proteinuria. Ultrasonography revealed minimal effusion in the lung and possibility of pulmonary sequestration. Ascites was absent. CECT chest was done (Figure 3) and final diagnosis of extralobar pulmonary sequestration was made. As the cause of pleural effusion was identified, karyotyping wasn't done.

Child's condition improved. NIPPV weaned off and child maintained saturation at room air. Child was transferred to nursery after 7 days. Cardiothoracic and vascular surgery (CTVS) consultation was done. The child was advised for follow up at 3 months for possible need of elective surgery.

DISCUSSION:

Pulmonary sequestration is a congenital lesion that consists of anomalous lung parenchyma that has own arterial supply with no connection to tracheobronchial tree. This rare abnormality has an incidence between 0.15 and 6.45% among all pulmonary malformations.[4] It is divided two intralobar (ILS) and extralobar pulmonary sequestration (ELS). Intralobar sequestration is contained within normal lung parenchyma with venous drainage to pulmonary vein whereas extralobar is separated from normal lung and has its own visceral pleura and has venous drainage to systemic vein.[5]

ELS accounts for only 25-15% of all sequestration.[6]Most ELS present in the first 6 months of life with one-quarter of babies presenting shortly after birth with respiratory distress or feeding difficulties.[7] Slightly older children present with respiratory symptoms and occasionally with congestive cardiac failure. It can be seen on fetal ultrasound as early as 18 weeks of gestation as a well-defined triangular echogenic mass in the lower chest or the suprarenal region of the abdomen.[8]

Neonatal pleural effusion secondary to ELS has rarely been reported in the literature. Earlier, Bibek AJ et al, Dresler S, Horowitz RN, J Lukaya et al have reported this unusual clinical condition.[3,9,10,11] Neonates in those case reports were born premature at or before 34 weeks of gestation. All of them developed respiratory distress immediately at birth and required vigorous resuscitation at delivery room. However, none could be saved and diagnosis was made based on the post mortem findings. In contrast to the earlier case reports, the neonate in our case was delivered late pretem at 36+2 WOG. Baby developed respiratory distress but did not require vigorous resuscitation. Similarly, thoracostomy to drain pleural fluid was done immediately which might have led to a favorable prognosis in contrary to other cases. Diagnosis on our neonate was done based on ultrasonography and contrast enhanced CT scan findings. Baby was discharged after improvement. Anomaly in our case was left sided, which was consistent with all other reported neonates.

It has been hypothesized that the obstruction to the lymphatic drainage of sequestration might be responsible for pulmonary effusion and associated pulmonary hypoplasia. Unilateral massive pleural effusion should also be added to the list of unusual forms of pulmonary sequestration in the neonatal period.

Most pulmonary sequestration show a spontaneous partial or complete regression during the course of pregnancy and can be managed expectantly with excellent prognosis. However, in some cases there might be increase in size and may develop massive pleural effusions with mediastinal shifting.[12] Currently, intrafetal vascular laser ablation of the feeding vessel (VLA) is a recommended highly effective treatment option.[13] Those ELS detected postnatal if asymptomatic can either undergo surgical resection or transcatheter arterial embolization or observation without treatement.. However, symptomatic extralobar pulmonary sequestrati-

on should undergo surgical resection. Elective surgical excision is planned at 3 months of age for neonate reported.

Conclusion:

Pulmonary sequestration should be suspected as a possible differential diagnosis in a fetus with evidence of polyhydramnios and pleural effusion.

Conflict of Interest: None.

Acknowledgement: None

Consent:

Written informed consent was obtained from the parents to publish this report in accordance with the journal's patient consent policy.

Data avaibility statement:

Data will be provided by the corresponding author upon reasonable request

Images uploaded in the separate files.

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