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CASE REPORT

Integrative Pediatrics

Successful resolution of congenital idiopathic non-chylous pleural effusion in newborn

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Abstract

Pleural effusion accompanied by non-chylous presentation is a rare occurrence in newborn. Herein, we report a case of an otherwise healthy newborn who was diagnosed as a case of idiopathic non-chylous pleural effusion after evaluation for all possible explanations. The findings and management followed therein were consistent with case reports from other occurrences with respect to pleural effusion but with the additional event of a pneumothorax in the patient, our management needed extension. Such an instance of a rare disease in association with pneumothorax makes this case report a valuable asset to the scientific literature, contributing towards shedding knowledge in an otherwise scarce literature..

Keywords: *Congenital, idiopathic non-chylous pleural effusion, Newborn, Respiratory distress*

Abbreviations: *PICC: Peripherally inserted central catheters, NICU: Neonatal intensive care unit, BPD: Bronchopulmonary dysplasia, TPN: Total parenteral nutrition*

Introduction

Pleural effusions are rare in the neonatal period [1]. With an estimated incidence of 0.06 percent, it occurs as fluid build-up in the pleural space when the production exceeds absorption of pleural fluid [2]. The etiology leading to pleural effusion in a neonate is critical and relevant as it is associated with multiple congenital and acquired lesions. Hydrops fetalis, extravasation of Peripherally inserted central catheters (PICC) or umbilical venous catheter, chylothorax, para-pneumonic effusion, congestive heart failure remains one of the most common causes. Diagnostic chests tap for pleural fluid analysis is important for diagnosis and its consequent management [3]. Any effusion in neonates without obvious explanations is termed as Idiopathic neonatal pleural effusion [4]. With most cases involving effusion usually as chylous, a minority of the cases have had non-chylous findings.

It is critical that neonatologists and obstetricians should be aware of such condition, which can present immediately after birth and needs vigilant delivery room management. It is also worth mentioning that pleural effusions can be identified on an antenatal ultrasound scan so if observed, delivery of the baby should be properly planned with neonatologist and done at a tertiary care center. Idiopathic non-chylous pleural effusion is a diagnosis of exclusion so all the possible causes of pleural effusions should be ruled out prior by the help of pleural fluid biochemistry and triglyceride levels. Echocardiography and karyotyping are also indicated. We herein report a case of pleural effusion in a newborn, which was non-chylous, and there were no other possible explanations.

Case report

A gravid 28-years old healthy female presented at 37-weeks of gestation with labor pains. Only one antenatal ultrasound scan was available which was performed at 30-weeks of gestation and was unremarkable. Cesarean section was done, and a baby boy was born with immediate cry and 8 and 9 Apgar score at 1 and 5 minutes respectively. Soon after birth, baby started to grunt and developed tachypnea with a respiratory rate of 70 breaths per minute. Oxygen saturations were 90% on room air after half an hour of birth. There was no dysmorphism or any other congenital abnormality. Baby had obvious signs of respiratory distress with subcostal recessions and nasal flaring. On auscultation bilateral air entry was decreased with no added sounds. There was no murmur and no signs of cardiac failure. Abdomen was soft with no sign of fluid in the abdominal cavity.

Baby was shifted to neonatal intensive care unit (NICU) nursery and oxygen inhalation along with workup for sepsis was done. A chest x-ray was also performed which demonstrated right sided pleural effusion and left sided pneumothorax as shown in figure 1, for which needle thoracotomy was done urgently and a chest tube was inserted, and pleural fluid was drained. Around 200ml of fluid was obtained. Ultrasound chest was done which showed residual fluid of around 50ml (after chest tube was inserted) and no locations were identified.

Pleural fluid was collected, and it was yellow in color. Pleural fluid biochemistry showed proteins of 2.53g/dL, glucose of 64mg/dL, LDH 257U/L, and specific gravity 1.022. Microscopic examination showed Red blood cells 300/mm^3 and white blood cells 3150/mm^3

with lymphocytic predominance of 95%. Triglyceride levels in pleural fluid were 54mg/dL and cholesterol levels were 65mg/dL, which shows that the fluid was non-chylous. Blood and pleural fluid cultures showed no growth. Ultrasound abdomen showed no fluid collection in the abdomen. Echocardiography was done to rule out cardiac failure which was also normal. Karyotyping was not performed.

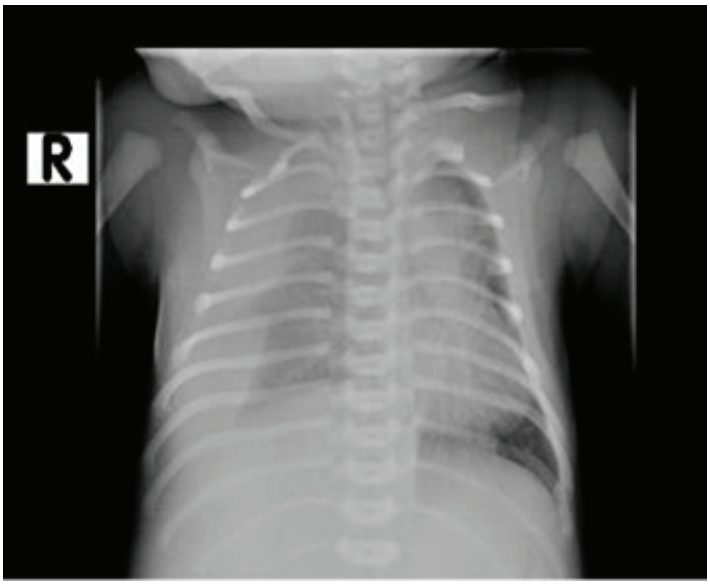

Figure 1. *Right sided pleural effusion*

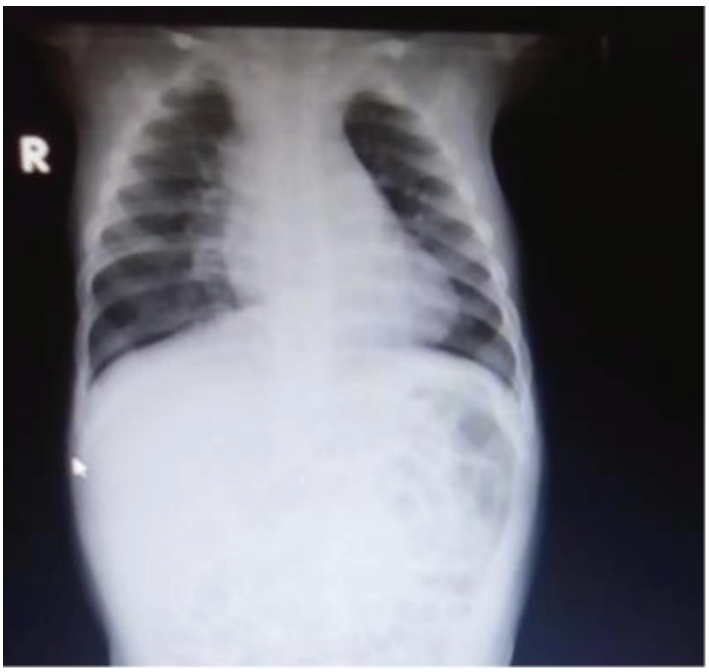

Figure 2. *Resolution of pleural effusion*

Child's general condition improved, and oxygen requirement decreased after chest tube insertion. Repeated chest x-ray was normal. Baby remained in hospital for around 8 days and was discharged in a stable condition without any oxygen support with resolved pleural effusion as shown in figure 2. At outpatient follow up the baby was doing well at 3 months of age. For the purpose of publication, informed consent in written form was obtained from the parent of this patient.

Discussion

Accumulation of fluid in pleural space of lungs is termed pleural effusion. A fine balance between the fluid producing visceral pleura and fluid absorbing lymphatics of parietal pleura maintains the volume of fluid in pleural space. If the fluid is produced in excess or the absorption is decreased, then it is collected in the pleural space. Causes of such occurrence in newborn include congenital, non-immune and immune hydrops, secondary to leaky capillaries, iatrogenic injury to thoracic duct leading to collection of chyle, Down and Turner syndromes, pneumonia and wet lung syndrome. Having no apparent cause, this case was diagnosed as idiopathic pleural effusion [3-5]. Neonatal idiopathic pleural effusion are usually chylous and once accumulated, it may turn chylous after administration of external fat feeds. Previous reports describe association of serous or non-chylous congenital pleural effusion with primary lymphangiectasia, congenital cystic chromosomal anomalies, adenoid malformation, bronchopulmonary dysplasia (BPD), chest wall hamartoma, diaphragmatic hernia, and pulmonary vein atresia [6-8]. Occurrence of idiopathic non-chylous effusion has been very rare similar to our report [4-9].

Pleural effusions are quantified by the help of ultrasound scan as: small (< 10 mm), moderate (10-30 mm) and large (> 30 mm), measured at the posterior pleural costophrenic recess with the patient in supine position [10]. Buttiker et al defines chylothorax against a criterion as that pleural fluid with an absolute white cell count > 1,000 cells/μL and a lymphocyte fraction > 80% [11]. Chylous pleural fluid should also report triglycerides to be more than 110 mg/dL (provided there was minimal fat enteral intake). Triglyceride levels in pleural fluid of our case were 54mg/dL and cholesterol levels were 65mg/dL, which shows that the fluid was non-chylous. Even after milk feeding and Total parenteral nutrition (TPN), this case is proven as a non-chylous effusion.

Pleural effusion can present with asphyxia and respiratory distress in newborns ranging from mild to severe forms [12]. Delivery room management should anticipate the needs of the most severely affected patients, which may include endotracheal intubation, positive pressure ventilation, and evacuation of the pleural effusion(s) by thoracentesis. These neonates are at risk for pneumothorax or pneumo-mediastinum because of pulmonary hypoplasia and poor lung compliance [3]. If pleural effusion is presented before 32-weeks of gestation, there is high mortality risk in these preterm babies [13]. Bilateral pleural effusions can lead to pulmonary hypoplasia, as lungs are unable to expend. Risk of chronic dependency on supportive oxygen is linked with effusions persisting beyond 3 days after birth [14]. Gold standard management incorporates performing thoracocentesis with subsequent intercostal drain. Accompanying the mentioned management, administration of antibiotics is continued till an infectious etiology is excluded on culture [1].

Idiopathic non-chylous neonatal pleural effusion is a rare entity, therefore it's prompt diagnosis and management can be lifesaving specially in cases of moderate to severe respiratory distress situations. It is vital to provide proper antenatal care, planning of delivery if possible, in tertiary center, so the adequate

post-natal care and resuscitation can be well anticipated for such cases. Further studies need to be conducted to assess long-term outcome of neonates with congenital chylothorax including neurodevelopmental follow-up and pulmonary function testing.

Ethical approval and consent to participate

Written informed consent was obtained from the parents of the patient for the case details to be used for any publication.

Consent to publish

Written informed consent to publish was obtained from the parents of the patient for publication of this case report in a journal as well for other study purposes.

Availability of data and material

Case details are not publicly available because the data is patient medical records but are available from the corresponding author on reasonable request

Author's contribution

SK, AM, OI and MAQ were involved in literature search of topic, medical record review, introduction design draft, critical revision and interpretation of data, and final approval and agreement. OI, MAQ, MM and SBA were involved in discussion design, medical record review, critical revision and review, and final approval and agreement.AL and KA were involved in critical revision and editing of final manuscript, and final approval and agreement. All authors fulfill the criteria and have contributed immensely to the final production of this manuscript. All authors read and approved the final manuscript.

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