EUROPEAN ACADEMIC RESEARCH
Vol. VII, Issue 9/ December 2019

Impact Factor: 3.4546 (UIF)
DRJI Value: 5.9 (B+)

ISSN 2286-4822
www.euacademic.org

Idiopathic Congenital Non-Chylous Pleural Effusion in a Newborn: A case report

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Abstract

Spontaneous or idiopathic pleural neonatal effusion is a rare disease. This condition causes neonatal respiratory distress. In this article, we will discuss a rare case involving a child diagnosed with idiopathic congenital non-chylous effusion. This was recorded for the first time in our clinic. The differential diagnosis at the beginning was made with congenital chylothorax as the main cause of congenital pleural effusion. After the examination, effusion was not chylous and there were no signs of a clear etiology nor related diseases or associated syndromes causing pleural effusion. The patient presented respiratory distress after birth and required mechanical ventilation as well as cardiopulmonary resuscitation. Treatment was done with thoracocentesis and the prognosis was successful. This article highlights a case with a rare disease and aims to underline the importance of early recognition of this pathology, the effective intervention to reduce mortality and a better understanding and isolation of the disease outcomes.

Key words: Idiopathic Pleural Effusion, Congenital, Non-Chylous, Respiratory Distress, Newborn

INTRODUCTION

Pleural effusion is the accumulation of fluid in the pleural space, found between the parietal pleura of the chest wall and the visceral pleura of the lung. Pleural effusion may occur at any time, antenatal, at birth or postnatal stage.[1]

Spontaneous or idiopathic pleural neonatal effusion is defined as the presence of the effusion in a newborn under 30 days of age [2,7,9].The disease is rare, without any obvious explanation and occurs in 1 to 12,000 up to 1 to 15,000 pregnancies[3,6].Pleural effusion can be congenital or acquired[4].Pleural effusion may be isolated or with anatomical malformations, chromosomal abnormalities and with genetic syndromes[5].The content of pleural fluid is chylous, but in a small number of cases it may be non-chylous[6].Chylotorax is the most likely cause in the neonatal period of pleural effusion[1,7].Pleural effusion is diagnosed via chest radiography and ultrasound.Idiopathic congenital pleural effusion is an exclusionary diagnosis and should be established after concomitant abnormalities are excluded, such as congenital diaphragmatic hernia, mediastinal hernia, congenital infections[5].This article outlines a rare case of a newborn with idiopathic pleural effusion non-chylous with severe respiratory failure.

PATIENT PRESENTATION

A female newborn was antenatal diagnosed through ultrasound with pleural effusion on the 34 week of pregnancy. The neonate was born on the 38th week. She weighed 3,100 grams, with a Apgar 4/3 on the 1/5 minute, with a severe respiratory failure, generalized cyanosis and an auscultation with weakened respiration of the right lung. The baby had tachypnea with respiratory rate 78/min and cardiac rate 190/min. In these conditions the baby was immediately intubated after birth and rushed to ICU for further treatment. During examination there was no sign of facial dysmorphia and neither hydrops. An urgent Chest X-ray was done showing an evident opacification of the right lung.The baby was punctuated in the 4th intercostal space, a 220ml yellow to orange pleural fluid was removed and a thoracic drain was inserted.. Analysis of pleural liquid showed a 2.3 g/L proteins level; 0.531 mmol/L triglycerides level; 0.984 mmol/Lcholesterol level and

the number of leukocytes ≤ 500 /mm3 reflecting transudate. Growth of microorganisms in liquid culture was not observed and the level of procalcitonine and inflammatory Protein-C in blood was on the normal range. Cardiac echocardiography was normal for congenital anomaly. CT-Thorax was done to exclude mediastinum and thoracic duct anomaly or tumor. Karyotype analysis resulted normal, i.e. 46XX. After 24 hour from thoracic drain insertion, an expansion of the right lung was noted and the clinical signs improved significantly. On the second day, a low level of 50 ml pleural fluid was removed. On the third day feeding started and there were no signs of leak or augmentation of pleural fluids and no change of color wasnoted. The baby was extubated on the fifth day. After ten days, the baby was discharged in good conditions.

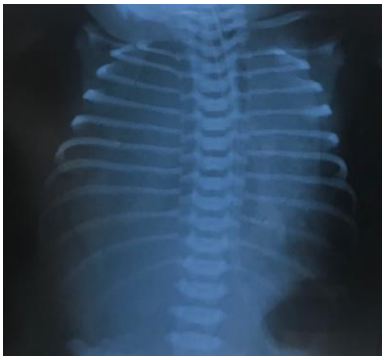

Fig 1.Chest X-ray showing white opacity of the right hemitorax.

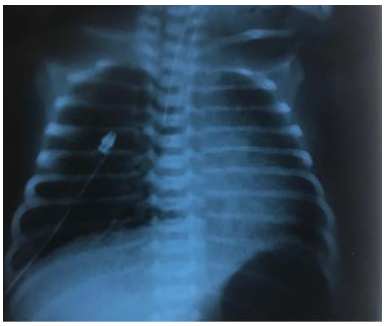

Fig 2. Chest X-ray after tube placement and fluid drainage

DISCUSSION

Pleural neonatal effusion can be manifested as a result of different pathologies or without any possible justifiable cause.Pleural neonatal

effusion is divided in two groups: congenital and acquired. Pleural effusion, because of the congenital pathology, counts nearly 32% of the cases, while 68% is as a result of acquired pathologies [4]. The causes of congenital pleural effusion are numerous, including chylothorax, immune and non-immune hydrops, part of Down syndrome, Turner, Noonan, congenital pneumonia, cardiac congenital anomaly and other [7,8].

Spontaneous or idiopathic neonatal pleural effusion is defined as any effusion in a newborn of age less than 30 days, without any obvious etiology [2,7,9].

Different researchers report that pleural effusion is serous, non-chylous, and is associated with Down syndrome[6].In our case, pleural effusion is non-chylous, but without any cause found by examinations. The primary limitation to the generalization of the result and outcome of the disease represents a first in our experience in relation to this pathology. Limited reports and analysis of this disease also represent an obstacle in this area.

In most cases, idiopathic neonatal pleural effusion is chylous[7].Chylous fluid has a milky color and diagnostic criteria for chylotorax are: levels of triglycerides more than 1.24 mmol/L,a cholesterol range of 1.68-5.69 mmol/L, leukocytosis and absolute cell account more than 1000/ μL with fraction of the lymphocytes more than 80%[5 ,10,11].

Clinical conditions of pleural neonatal effusion can be asymptomatic or with moderate signs of respiratory distress up to severe respiratory failure that need resuscitation and mechanical ventilation as in our case report. In most cases, simple effusion changes to chylous after starting of external fat feeds[2,6,12,7].In our case, the effusion remained non-chylous after feeding. In cases involving severe respiratory failure with large pleural effusion, in order to expand the chest wall it is not needed only early intubation and mechanical ventilation, but it is necessary an early management with thoracocentesis. To make a quick and a good assessment of the diagnosis of severe respiratory distress after birth, it is necessary an urgent chest X-ray. The ultrasound is very helpful to make an antenatal diagnosis of congenital pleural effusion. Diagnosis of pleural effusion made antenatal before 32 weeks, have a mortality rate as high as 55%[7,13,14,]. For a precise diagnosis of etiology it is mandatory the thoracocentesis or thoracostomy to analyse pleural

liquid for levels of proteins, level of lactate dehydrogenase, numbers of cells and differential, microbiological analysis and the culture of liquid.[1].CT scan of thorax has a supplementary role in differentiation of pleural effusion that can present radiologically as lucent or opaque hemithorax with contralateral mediastinal shift[9].CT scan is also necessary to exclude other abnormalities of mediastinum and thoracic duct or tumor.[9].In our case was not evidenced any anomaly with CT scan. Management of pleural effusion includes mandatory thoracentesis and intercostal tub insertion [7,15].In our case was observed a good improvement of clinical signs within 24 hours after the insertion of intercostals tub. The neonate with hypoplastic lungs has a bad prognosis and it is often noticed with bilateral pleural effusion. Our case was non-chylous and it was necessary only thoracentesis and the treatment with antibiotic was given until profile sepsis screening was excluded.

CONCLUSION

Respiratory distress remains one of main manifestations of neonatal pathologies. Idiopathic pleural neonatal effusion is a rare cause of respiratory distress that can be presented in the context of severe respiratory failure and may result life threatening. Hence, it is essential to have awareness on the existence of this pathology as idiopathic phenomena[16]. Early recognition of this disease is important for a quick and accurately diagnosis, efficiently management and a full recovery.

Grant Support
None.

Conflict of interest
The authors declare that they have no competing interests.

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