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RESEARCH ARTICLE

CONGENITAL PULMONARY ADENOMATOID MALFORMATION TYPE I: A CASE REPORT WITH CLASSICAL HRCT FINDINGS

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ABSTRACT

Congenital cystic adenomatoid malformation (CCAM) is usually observed in neonates because of respiratory distress and may occasionally be observed in older children or adults with recurrent infection 1. This malformation consists of adenomatoid proliferation of bronchioles that form cysts instead of normal alveoli. We report a case of a 11 days old neonate who presented with complaints of severe and persistent respiratory distress. Complete blood investigations, sequential chest radiographs and HRCT thorax of the neonate were performed. This study highlights the role of HRCT in reaching the diagnosis and further guiding the treatment.

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INTRODUCTION

A 11 days old male neonate born of normal vaginal delivery presented to the paediatric emergency of SSG hospital, Vadodara with complaints of severe persistent tachypneoa since birth. No prior antenatal scan had been performed. The baby was afebrile. Chest radiographs showed hyperlucency with absent broncho-vascular markings involving whole of the right lung field with mediastinal and tracheal shift to the contralateral side. Rest of the visualised lung parenchyma was normal with no evidence of pleural effusion on other aide. Proper respiratory support plus oxygen therapy was given to the patient but the symptoms kept on deteriorating. Blood investigations were within normal limits-hemoglobin of 17gm/dL, total leukocyte count of 4500/ cm3, differential count of polymorphs 62%, lymphocyte 36%, eosinophil 2%, and platelet count of 3.02 lakhs with erythrocyte sedimentation rate of 38mm (1st hour). Liver function tests and kidney function tests were essentially normaland cultures were negative. Therefore, HRCT chest was performed to evaluate the cause of the respiratory distress. HRCT chest showed multiple, variable sized, air filled cystic lesions confined to the lower lobe of right lung with mediastinal shift to the contralateral side and partial volume loss of the rest of the right lobe secondary to pressure compression.

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DISCUSSION

Congenital pulmonary adenomatoid malformations, previously known as congenital cystic adenomatoid malformation are rare lesions presenting with neonatal respiratory distress. It was divided into three types depending on the size of the cysts. The typical imaging findings of type 1 CPAM are one or more large air-filled cystic lesions [Figure 2, 3] (Paterson, 2005; Stocker, 2002 and Yıkılmaz, 2007). Multiple cystic lesions can cause mediastinal shift and herniation of the affected lobe (Paterson *et al*., 2005).

Type 2 CPAM usually consists of an air-filled multicystic mass or focal area of consolidations. Type 3 CPAM tends to be seen as homogeneous soft tissue density mass due to microscopic cysts that can be identified only at histologic evaluation. Type 4 CPAM consists of large air-filled cysts of distal acinar origin (Paterson, 2005; Stocker, 2002 and Yıkılmaz, 2007). Differential diagnosis of Type 1 or Type 2 CPAM in a neonate includes congenital diaphragmatic hernia, pulmonary sequestration, bronchogenic cyst, CLE, and other bronchopulmonary foregut malformations (Paterson, 2005; Kim, 1997). Our patient was taken up for surgery and right lower lobectomy was done. Histopathological examination showed multiple variable sized air and fluid filled cysts.

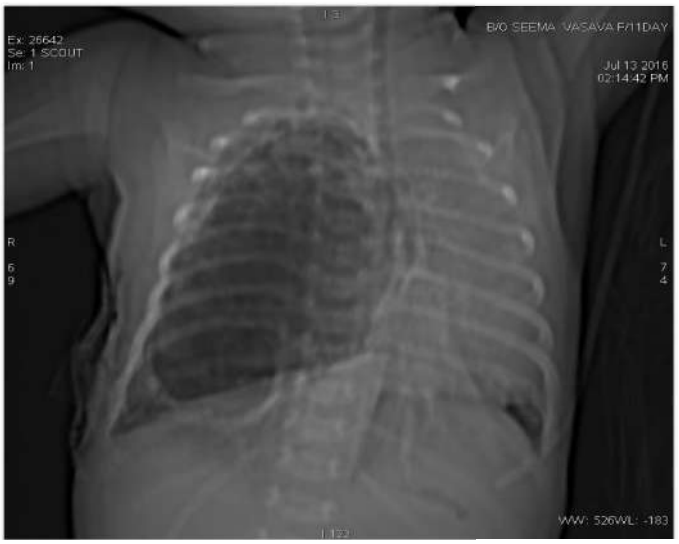

Fig. 1. Scout topogram showing a large well defined hyperlucency with absent broncho-vascular markings occupying whole of the right lung field with shift of mediastinum and trachea to the contralateral side. Rest of the visualised lung parenchyma appeared normal. No evidence of pleural effusion of either side. Visualised bony thorax appears normal

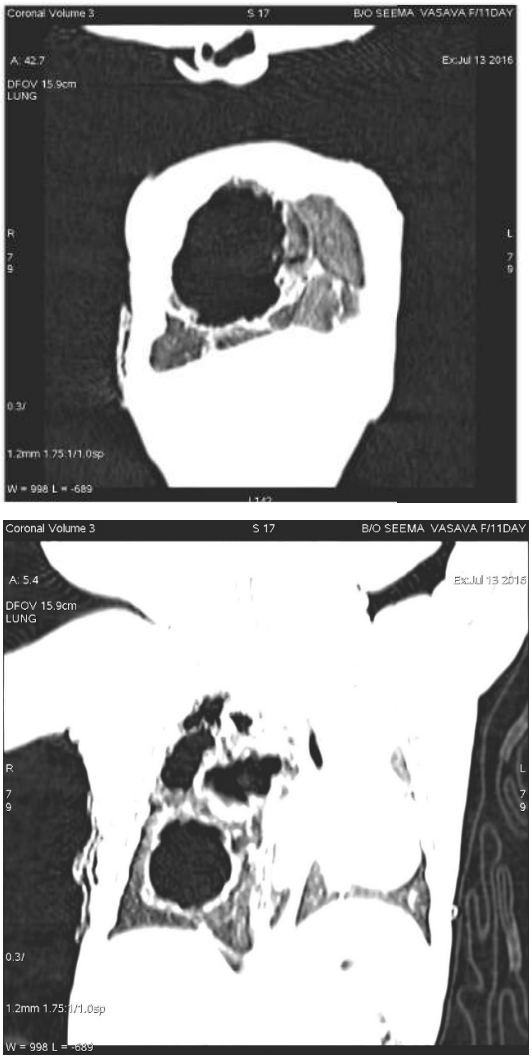

Fig. 2 a,b. Coronal reformatted images in the lung window shows multiple variable sized, air containing cytic lesions with thick walls confined to the lower lobe of right lung causing pressure effects with partial volume loss of the rest of the lobes of bilateral lung fields. The right major and minor fissure, mediastinum and trachea are pushed to the opposite side

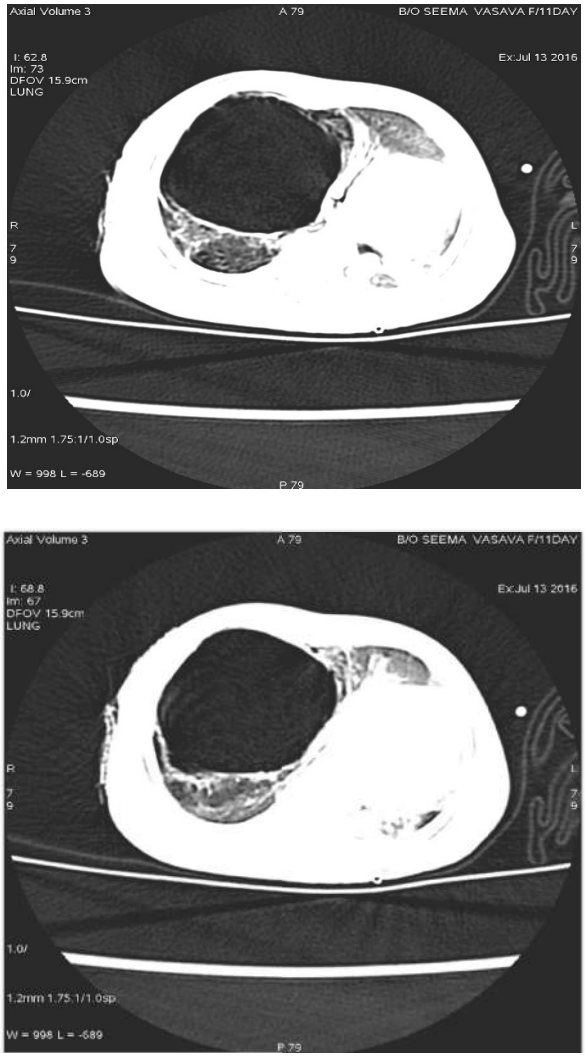

Fig. 3 a,b. Axial images in the lung window shows large round air containing cytic lesions with thick walls confined to the lower lobe of right lung with displacement of the right major and minor fissure

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