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Bronchogenic Cyst: a rare cause of respiratory distress in neonate - A Case Report

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ABSTRACT

Bronchogenic cyst is an unusual congenital pulmonary malformation which may occur at various sites of tracheobronchial tree as well as unusual ectopic site like cervical, scapular region, pericardium and abdomen. Newborn with large cyst can develop respiratory distress, cyanosis and feeding difficulty. A 37 day old male infant presented with failure to thrive, feeding difficulty, respiratory distress and cyanosis. His chest radiograph was suggestive of shift of mediastinum and heart with hyperinflatted left lung. CT thorax showed minimal enhancing cystic lesion posterior to trachea and its bifurcation on right side, it was compressing the right main bronchus. The cyst was successfully surgically excised which was adhering to carina and intrabronchial extension and its histopathological finding confirmed the bronchogenic cyst. Though, bronchogenic cyst is rare cause of respiratory distress, cyanosis, and failure to thrive and feeding difficulty, it should be considered one of the differential diagnoses.

Keywords: Bronchogenic cyst, Congenital pulmonary malformation, Neonatal respiratory distress

Congenital Pulmonary Malformations (CPMs) are a group of lung abnormalities affecting the airways, parenchyma, and vasculature. Congenital cystic diseases of the lung encompass a spectrum of anomalies ranging from congenital cystic adenomatoid malformation, brochogenic cyst, bronchopulmonary sequestration, congenital lobar emphysema, pleuropulmonary blastoma. Bronchogenic cyst (BC) is an unusual congenital lung development anomaly, with prevalence rate of 1 per 68000 births [1]. BC develops from abnormal budding of tracheal diverticula and can be classified into three categories in relation to the trachea as paratracheally, intraluminally and intramurally. It can be detected as incidental finding on chest radiograph and account for 10% of the mediastinal masses in children but newborn with large cysts can develop respiratory distress, cyanosis and feeding difficulty [2]. We report a neonate with bronchogenic cyst at carina and right bronchus with respiratory distress.

CASE REPORT

A 37 days old male infant was admitted in emergency department with history of respiratory difficulty and bluish discoloration of hands, feet and lips with excessive crying. On further inquiry, history of recurrent cough, cold without fever since 20 days was revealed. The infant was hospitalized and evaluated by primary care physician as a congenital heart disease and treated as congestive cardiac failure but he did not respond to treatment, so referred to our institute. He was born at 38 weeks of gestation of 23 years old primi mother, through vaginal delivery with birth weight of 2.5 kg with adequate antenatal care, without any history of maternal drug intake or radiation exposure, with uncomplicated postnatal period. He had no history of congenital malformation in family.

On physical examination, he had signs of failure to thrive (weight 1.9kg), heart rate 152/min, respiratory rate 86/min with signs of respiratory distress, duskiness of lips, hands and feet (SpO2 80% without oxygen and 97% with oxygen). He was irritable, normothermic with normal capillary refill time, and no sacral edema. He had decreased air entry on right hemithorax with fine crepitation and distinct heart sound on right hemithorax with palpable liver.

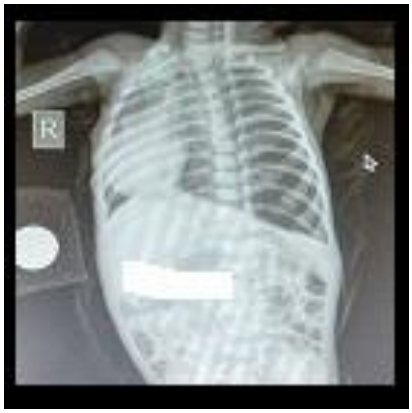

Figure 1: Chest X-ray showing right shift of mediastinum and heart and hyperinflation of the left lung

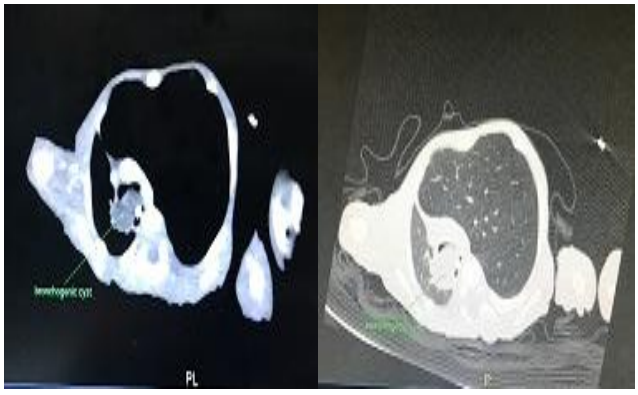

Figure 2A & 2B: CT thorax showing cystic mass

Blood investigations including complete hemogram, platelet count, micro ESR, blood sugar, serum calcium, electrolytes, and renal function test were within normal limit. C-reactive protein and blood culture was negative. Blood gas analysis was suggestive of mild respiratory acidosis. Chest X-ray showed shift of mediastinum and heart to right hemithorax and hyperinflatted left lung (**Fig. 1**). A two dimensional echocardiography was within normal limit. Multislice spiral Computed Tomography (CT) revealed well defined minimally enhancing cystic lesion of average attenuation, posterior to trachea and its bifurcation on right side and anterior to esophagus extending from T3 to T6 vertebrae causing compression of right main bronchus resulting in partial collapse of right lung field and hyperinflation of left lung with mediastinal shift to right (**Fig. 2A & 2B**).

In view of clinical and CT finding, a diagnosis of bronchogenic cyst was made and excision of the cyst was planned. By mid axillary incision, right thoracotomy revealed a bronchogenic cyst of about 1 cm in diameter in the carina slightly to right side below the bronchus which was adhering to carina and right bronchus. For separation of cyst, carina and bronchus was cut open, there was intrabronchial extension of the cyst compressing the carina. The cyst was cut open and about 1ml of whitish jelly like substance was collected out of the cyst. Left side intercostals drain was inserted for improving intraoperative ventilation. The cyst was excised, carina and right bronchus were closed and mediastinum was exposed to rule out any cystic lesion. Histopathological examination showed fibromuscular wall lined by pseudostratified ciliated columnar epithelium with dilated blood vessel and mucous gland (**Fig. 3A & 3B).**

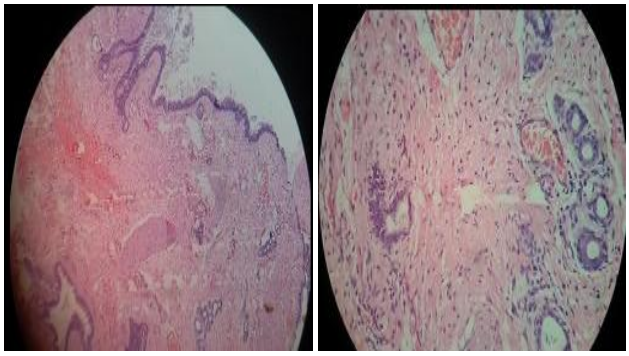

Figure 3A & 3B: Histo-pathological changes of bronchogenic cyst

DISCUSSION

During the third week of embryonic development, the dorsal portion of the primitive foregut lengthens to form the esophagus, and the ventral portion undergoes differentiation to form the tracheobronchial tree. Insufficiency in the process of growing of the ventral foregut will give rise to BC. Bronchogenic cyst develops from abnormal budding of the tracheal diverticula [3]. Though antenatal discovery of BC is rare and location is generally intrathoracic, with improved resolution of foetal sonography and Doppler studies many of these lesions are

detected "in utero". Most of the bronchogenic cyst originates in the mediastinum, while 15%-20% occur in the lung parenchyma and most of them in the lower lobes [4]. They may occur at paratracheal areas, above the tracheal bifurcation, at the level of carina, paraesophageal area, hilar (in which the cyst is attached to one of the main bronchi), suprasternal notch and may rarely from lobar bronchi. Rarely when buds detach and migrate to ectopic sites, cyst may be found in cervical, scapular region, pericardial and abdominal [5-7].

Although some BC are asymptomatic and are the incidental finding upon radiography, most of them are symptomatic and the most frequent symptoms are cough, fever, wheeze, and dysponea[4]. Newborns with large cysts can develop respiratory distress, cyanosis and feeding difficulty. Compression of the trachea and respiratory arrest, wheeze and recurrent pneumonia in BC is not rare. Our case has had similar presentation mimicking congenital heart disease. Life threatening complications like recurrent severe pneumonia, respiratory arrest and cardiac arrest are reported in literature [8]. BC is also associated with various congenital anomalies of heart, chest wall and diaphragm. Imperatori A et al [9] and Kamata T et al [10] observed bronchogenic cyst with pericardial defect in adult. There is a small risk of malignant transformation within the cyst; bronchoalveolar cell carcinoma and neuroblastoma are reported with bronchogenic cyst [2].

The postnatal diagnosis of BC may be suspected on chest radiographs but ineffective for accurate preoperative diagnosis. CT thorax is valuable in demonstrating the size and shape of the cyst and in determining its position in relation to other structure. Bronchogenic cysts are thin walled and may contain blood vessels, mucous gland, smooth muscle, elastic tissue and cartilage. Making preoperative diagnosis of BC can be difficult. Treatment of asymptomatic BC is controversial but risk of infection, bleeding, compressive symptoms like respiratory distress and chance of development of carcinoma need surgical excision in order to avoid complication. Transtracheal and percutanous cyst aspiration have been proposed as alternative to operation, but these methods are not widely accepted because of possible cyst recurrence [4]. Excision of cyst is the definitive treatment in symptomatic cases. A definitive diagnosis is not always possible preoperatively but it is recommended that surgical resection should be done in all suspected BC in operable candidate.

CONCLUSION

Usually congenital heart diseases with congestive cardiac failure, respiratory distress syndrome, muconeium aspiration syndrome and lactation failure are presented with respiratory distress, failure to thrive and repeated chest infection. Though, bronchogenic cyst is a rare congenital pulmonary malformation in children, it should be suspected in a neonate with respiratory distress.

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