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Rare Dual Congenital Anomalies : Bronchogenic Cyst and Atrial Septal Defect

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ABSTRACT

We report an extremely rare case of a Bronchogenic Cyst incidentally identified via contrast enhanced computed tomography in a one and half-month-old male infant, a known case of Ostium Secundum Atrial Septal Defect and mild pulmonary hypertension. The patient was evaluated in detail and cystic mass resembling a Bronchogenic Cyst was found over lower paratracheal mediastinal space. The patient was planned for excision of the posterior mediastinal lesion using a right limited postero-lateral thoracotomy incision and complete excision was done and the mass was pathologically confirmed to be a Bronchogenic Cyst. This case is one of the few rare cases of infants with acyanotic congenital heart defect that were incidentally found to have a Bronchogenic Cyst. This case to our knowledge is one of the youngest patients yet and also highlights the importance of identifying rare causes like these amongst differentials of cough and wheeze responding poorly to regular treatment.

Keywords: *Bronchogenic cyst, Paediatric cardiac surgery, Paediatric congenital heart disease, Mediastinal mass*

1. INTRODUCTION

Mediastinal masses are extremely rare, asymptomatic and usually detected accidentally. Bronchogenic cysts consist of approximately 6%15% of the mediastinal tumours. Bronchogenic cysts are developmental anomalies occurring during embryogenesis. Congenital cystic lesions of the lung are usually benign producing no to minimal symptoms. However, some may present with life-threatening conditions. Considering the possible risk of symptomatic compression, the topography of the cyst is more important than the volume. Bronchogenic cysts are usually incidental findings found during evaluation for other congenital conditions of the new-born. Here, we present a case of a one and half year-old infant who initially presented with Ostium Secundum Atrial Septal Defect and mild pulmonary hypertension which was worsening, on further detailed evaluation incidental finding of a cystic mass resembling bronchogenic cyst was found arising from the lower paratracheal mediastinal space and the patient was planned for excision of the posterior mediastinal lesion which was later pathologically confirmed as a bronchogenic cyst. As per our knowledge this is one of the youngest patients to present with an incidental bronchogenic cyst in association with an Ostium Secundum Atrial Septal Defect and pulmonary hypertension which was treated successfully with no post-operative event. Therefore, it is extremely important to not exclude mediastinal masses as a differential in newborn infants presenting with a history of worsening congenital heart disease or wheeze and stridor. [(1)]

2. CASE REPORT

We present a case of a one and half monthold male child, a known case of acyanotic congenital heart disease (CHD). He was brought to our outpatient clinic with the primary diagnosis of atrial septal defect (ASD) and mild pulmonary arterial hypertension (PAH) which was managed medically. The patient presented a fortnight later to the emergency department of our institution with the chief complains of cough, fever and difficulty in breathing. He had dry cough associated with nasal blockage for three days which was followed by lowgrade, intermittent fever for 2 days which was relieved on taking medication. One day later, he developed difficulty in breathing and increased effort of breathing, excessive crying and irritability and reduced oral intake.

On admission respiratory rate was 66/min, subcostal retraction present, bilateral wheezing present, Spo2 was 96% on 4L Oxygen, As the respiratory distress worsened with altered sensorium and Arterial blood gas reports were suggestive of hypoxia, patient was kept on nasal continuous positive airway pressure (CPAP) and intubated later on. The patient reported negative on SARS CoV-2 analysis.

Initial chest X-ray findings revealed an incidental finding of a space-occupying lesion in the posterior mediastinum which was suggestive of a bronchogenic cyst (Figure 1). Confirmatory Contrastenhanced computed tomography (CECT) revealed a benign cystic right lower paratracheal space-occupying lesion (SOL); mostly likely suggestive of bronchogenic cyst and it caused distal segmental collapse/consolidation of the posterior segment of the right upper lobe (*Figure 2*). Consolidation involving superior segment of the left lower lobe was also visualized. Preoperative ECHO and Colour Doppler findings revealed a large Ostium Secundum ASD with a left-right shunt, mildly dilated right atrium and ventricle and mild PAH which was consistent with the initial diagnosis.

The patient was planned for the excision of posterior mediastinal lesion using a right limited postero-lateral thoracotomy incision (*Figure 3*). The excised mass was evaluated via histopathology and was consistent with the diagnosis of bronchogenic cyst. The patient was operated uneventfully and was shifted to Paediatric Intensive Care Unit, SKH. Patient was extubated postoperatively after 94 hours of total mechanical ventilation since intubation. Gradually patient was initiated on expressed breast milk (EBM) and was coping well with full EBM feeds and was discharged in a stable and healthy condition with a healthy wound. General paediatric review for immunization, feeding etc was advised to the patient before discharge. Post-operative ECHO and Colour Doppler revealed mild-moderate tricuspid valve regurgitation, mild left atrial ventricle dilatation, moderate PAH and good bi-ventricular function without any pleural or pericardial effusion. Postoperative period was uneventful and the patient is thriving well.

DISCUSSION

Mediastinal masses like bronchogenic cyst are usually asymptomatic when developed in adults; however, in infants and neonates, these can be life threatening. Bronchogenic Cysts are extremely rare and are usually excluded amongst differentials of stridor, dysphagia or neck masses amongst the paediatric population [(2)]. A ventral foregut budding abnormality during embryonic development usually leads to such malformations. Bronchogenic cysts can be unilocular, spherical or oval in presentation and are lined by ciliated columnar epithelium and fragments of cartilage and smooth muscle in the walls [(2)]. 85% of bronchogenic cysts present as mediastinal masses comprising of paratracheal, hilar and subcarinal space. 12% of them form the intra-parenchymal space (lower lobes) and are rarely found within the neck, pericardial or abdominal regions [(3)].

Bronchogenic cysts are usually intrathoracic in location and symptoms are attributed to the compression or displacement of adjacent structures (main stem bronchi, distal trachea, oesophagus, right pulmonary artery, and left atrium). Lazar et al reported that bronchogenic cysts usually prove lethal amongst infants and neonates owing to their softer, malleable tracheobronchial tree than older patients. Our patient developed sudden respiratory distress and was subsequently intubated and was in a critical condition with 94 hrs of ICU turnaround time [(2)]. Symptoms are usually in lieu of the cyst compressing neighbouring structures or the cyst ending up getting infected. 70.8% of the bronchogenic cysts were symptomatic in nature as reported in a previous study by Ribet et al [(4)].

There are no specific CT or Magnetic Resonance Imaging (MRI) findings that can help differentiate bronchogenic cysts from other mediastinal lesions prior to the histopathological investigations [(5)]. However, some common findings can be visualized on regular radiological investigations. Chest radiographs are the usual initial investigation of choice which may exhibit fluid filled cysts as radio-opaque masses. Hypoechoicity is visualized on ultrasound imaging. CT (0-20 HU) depicts low attenuation and on MRI, T2 hyperintensity with variable T1 signals is reported for bronchogenic cysts.

Haemorrhagic cysts may return a T1 hyperintensity. Some cysts may develop a layer of calcium oxalate which presents as hyperdense and hyperattenuated lesions on plain radiographs and CT scans respectively [(3)]. Bronchogenic cyst was an incidental finding in our patient and the findings were confirmed on CCECT. Final diagnosis was confirmed after histopathological evaluation of the excised mass. We could conclude that CECT was the confirmatory investigation of choice to rule out differential of a mediastinal mass.

The first complete excision of a bronchogenic cyst was performed by Maier [(6)]. Complete excision is the treatment of choice as these lesions are extremely prone to get enlarged or infected. Surgical approaches like thoracotomy and video assisted thoracic surgery (VATS) are used in case of operable patients in view of absence of adherence to mediastinal organs [(7)]. Our patient was operated upon using a right limited postero-lateral thoracotomy approach and complete excision of bronchogenic cyst was achieved without an intra-operative event. Several newer techniques have been used for excision of these cysts like Imperatori et al emphasized the use of harmonic scalpel via thoracoscopic approach. This reduces the blood loss, duration of drainage and length of the VATS procedure with cost reduction as compared to an electrical cautery [(7)]. Several congenital abnormalities have been found associated with bronchogenic cyst, some of them are Patent Ductus Arteriosus (PDA), atrial septal defect, tetralogy of fallot, mitral stenosis, pulmonary sequestration and diaphragmatic hernia [(4)(7)].Our patient was a known case of ASD with mild PAH and was diagnosed incidentally with a mediastinal mass suggestive of bronchogenic cyst.

Though the initial symptoms of difficulty in breathing, excessive crying, irritability, dry cough, mild fever and reduced oral intake could be attributed to the congenital heart defect or development of pneumonia. Failure to respond to initial therapy

should strongly persuade us to go for additional evaluation for the root cause. Physicians should look for differentials that could present with pneumonia-like symptoms. Bronchogenic Cysts may mimic pneumonia symptoms and should be under consideration if the patient is not responding to usual therapy. Surgical excision of the cyst is the management of choice and should not be preceded by conservative management as it is time consuming and does not provide any favourable outcomes. Complete excision also helps prevent unexpected events and complications.

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