Calcified Bronchocele

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INTRODUCTION:
Congenital bronchial atresia, a rather uncommon congenital anomaly, consists of atresia or stenosis of a lobar, segmental or subsegmental bronchus at or near its origin with normal development of distal structures. Distal to the stenosis the bronchi may become filled with mucus to form a bronchocele. The contents of the dilated bronchus may vary from mucoid contents to inspissated material or very rarely the contents may get calcified. Chest roentgenograms, spiral CT and MRI features of congenital bronchial atresia with or without bronchocele have been described though at present, spiral CT is accepted as the most sensitive investigation in making the diagnosis. To the best of our knowledge very few reports of calcified bronchocele associated with congenital bronchial atresia have been described.

We report a case of calcified bronchocele associated with congenital bronchial atresia.

CASE REPORT:
An asymptomatic 28 year old male was referred for the evaluation of a branching, tubular opacity in the right lower lung field, detected on a routine chest roentgenogram. The patient denied any specific respiratory symptoms and there was no other relevant medical or surgical history. The patient was a nonsmoker. The physical examination was unremarkable. Routine blood and urine examinations were normal.

Evaluation of the chest roentgenogram revealed tubular, branching opacities in the right lower lung field surrounded by an area of hyperlucency. Computed Tomography (CT) scan was performed which revealed homogenous, tubular lobulated branching opacities, surrounded by hyperinflated, oligemic lung parenchyma in the right lower lung lobe. The CT absorption coefficients of the lesion ranged between 450 & 500HU and the opacities did not show any enhancement following intravenous contrast administration. There was no contralateral mediastinal shift or significant collapse of adjacent lobes. Fibreoptic bronchoscopy was performed which confirmed the diagnosis of bronchial atresia.

DISCUSSION:
Congenital bronchial atresia is seen most commonly in young, adult males. 50% of these patients are...
asymptomatic. The abnormality is usually discovered on a routine chest roentgenogram (1,2). Symptomatic patients may present with recurrent pneumonia, dyspnoea, cough, hemoptysis or rarely as neonatal respiratory distress. A localized area of reduced breath sounds is the most common finding in most patients reported in the literature (1) and wheezing is heard in patients with a history of asthma. The diagnosis is suspected by the radiographic finding of a juxtahilar mass surrounded by regional hypertranslucency (3). Additional radiographic features may include mucus filled bronchi (bronchocele) and branching opacities radiating from the hilum. Air trapping around the affected area that cannot be eliminated during expiration probably due to a check valve mechanism, results in an obstructive segmental or lobar emphysema. A CT scan confirms the above finding and is currently the most sensitive test available for demonstrating the features of congenital bronchial atresia and to rule out the presence of an obstructing endobronchial lesion (4). Spiral CT with multiplanar volumetric reconstruction is helpful in distinguishing a mucocele from vascular malformation (4). Other imaging techniques including bronchography and MRI, are either difficult to interpret or unable to define the regional hypertranslucency around the bronchocele that is well visualized by CT scan. Thoracic MRI when performed, reveals the bronchocele as a branching opacity displaying hyperintensity on both T1 Weighted and T2 Weighted images due to the high level of protein content. The impacted mucus in bronchoceles may get calcified (5) and then these lesions are depicted as signal voids on both T1W and T2W images (6). Arteriography is used to exclude the presence of pulmonary sequestration, if suspected.

Spiral CT is the examination of choice, not only to show all the components of the anomaly and to estimate the extent of air trapping but also for ruling out differential diagnosis such as bronchogenic cyst, bronchiectasis, aspergillosis, completely thrombosed arteriovenous malformation or pulmonary aneurysms and tumors.

The frequency of bronchial involvement is in the following order: left upper lobe 64%, left lower lobe 14% and right lower and middle lobes in only 8% (2).

Development of bronchial buds occurs between the 4th and 15th weeks of gestation (7). A vascular insult during this period may cause interruption of the process, resulting in fibrosis and atresia of the involved segment. The normal appearance of the lung parenchyma distal to the atretic segment is supportive of a late development of the atresia after bronchial development is completed. The high frequency of involvement of the left upper lobe supports the ischemic theory, since the left upper lobe is the area of embryonic instability that develops late in utero. Beside congenital bronchial atresia, bronchoceles are seen in cases of acquired bronchial obstruction secondary to allergic bronchopulmonary aspergillosis, malignant tumors and tuberculous bronchostenosis and can be confused with tuberculomas, hydatid cysts and solitary pulmonary nodules. However, distal hyperinflation is seen only in bronchial atresia, intralobar pulmonary sequestration and bronchogenic cysts (8). Radiography, CT and bronchoscopy usually help make the distinction among these differentials.

Pathologic findings in patients who underwent resection, revealed that the left upper lobe was the most common location. Grossly the involved segment or lobe is markedly emphysematous and free of anthracotic pigment because of the relative lack of regional ventilation. Cystic dilatation of the atretic bronchus is distinguished from bronchiectasis by the presence of normally branching and distally extending bronchi. The involved parenchyma shows alveolar hypoplasia, a probable consequence of decreased ventilation and perfusion. These alveoli however remain open by collateral air drift occurring by the interalveolar pores of Kohn (air drift) or the bronchoalveolar channels of Lambert or the interbronchiolar channels (1).

In conclusion, spiral CT scan appears to be the imaging
modality of choice to evaluate and diagnose patients with congenital bronchial atresia and bronchocele. Asymptomatic patients may be managed conservatively while patients presenting with complications like recurrent pneumonia are preferably treated surgically.

REFERENCES: