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Lung Agenesis:A Case Report

KEYWORDS: Pulmonary Agenesis, Left Lung Collapse, Bronchoscopy , Spirometry

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ABSTRACT Lung agenesis is an extremely rare congenital anomaly representing failure of development of primitive lung bud. The aetiology of this condition is unknown, though genetic factors, viral agents, dietary deficiency of vitamin A during pregnancy have been implicated. Left sided lung agenesis is more common and the subjects have a longer life expectancy than those with right sided agenesis. The onset of symptoms in pulmonary agenesis is remarkably variable. In many cases, presence of this anomaly usually comes to light during infancy because of recurrent chest infections, cardiopulmonary insufficiency or due to associated congenital anomalies depicting severe obstructive airway disease on spirometry. Often, diagnose incidentally during the radiography of chest.

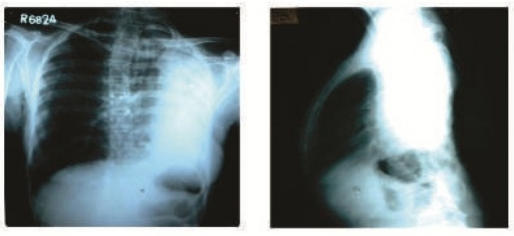
INTRODUCTION

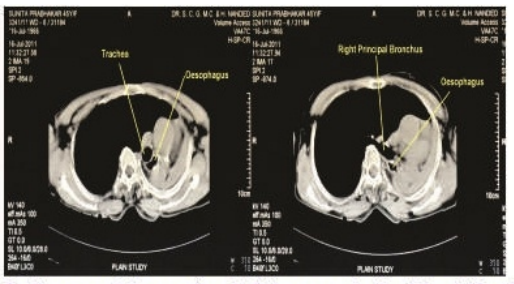
Agenesis of lung is an extremely rare anomaly representing failure of development of the Primitive lung bud. the condition was first discovered accidentally at the autopsy of an adult female.In 1673, by Dr. DePozze.[1] This condition is generally diagnosed in childhood. However, patients without any comorbid anomalies or patient with mild form of this disease can reach adulthood. It occurs approximately in 1 out of 100,000 births. No significant etiological factors have been found in patients thus genetic, teratogenicity, and mechanical factors have been thought to be responsible.[2]

CASE REPORT

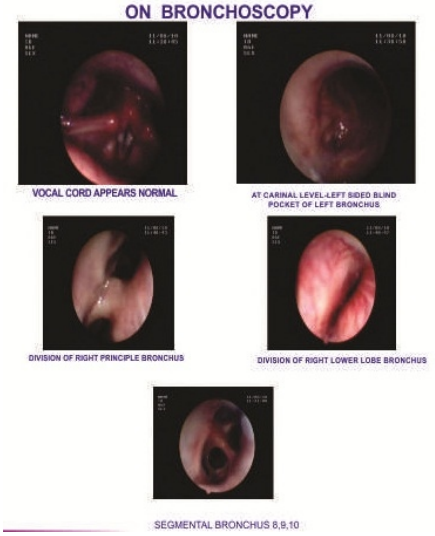
Fifty five years old female patients, house wife, came with complaints of Fever since 1 month, low grade which relieved after over the counter oral medications. Cough with expectoration since 1 month. Non-foul smelling not associated with postural or diunal variations. No significant occupational or family or personal history.

On general examination vitals stable. she was pale. b/1parotid and Submandibular bulge observed since childhood.





Collapse of the entire left lung noted with shift of heart and mediastinum to the left



Respiratory system

Upper respiratory tract normal.

Inspection: movement of left hemi thorax was diminished with apex impulse in left 5th1CS 3 cms lateral to midclavicular line. Palpation: inspectory finding confirmed Percussion: dull note on left hemi thorax with right side hyper resonant. Auscultation: respiratory sounds diminished in intensity on left with apical area tubular bronchial breathing with right side minimal rhonchi.

Chest x-ray and CT Scan: Collapse of the entire left lung noted with shift of heart and mediastinum to the left. Bronchoscopy: vocal cord Visualized normal with normal movement. Trachea visualized normal, left bronchus not visualized i.e. absent carina, right bronchus normal followed by anatomical disturbance – segmental anatomy disturbed. Pulmonary function test revealed severe obstructive airway disease with poor bronchodilator response with small airway involvement. Broncho-alveolar lavage report, inflammatory smear FNAC of parotid gland normal. So final diagnosis agenesis of left lung with compensatory right sided emphysema.

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DISCUSSION

Pulmonary agenesis means undeveloped pulmonary vessels, bronchi and parenchyma. Pulmonary aplasia (agenesis) is thought to result from the negative effects that occur on the 4th week of fetal life. [3]Although its etiology is not fully understood, vitamin A or folic acid deficiency or the use of salicylates may be responsible for it its incidence in male and female and the occurrence of anomaly in the right or left lung are about the same.

Schneider classified agenesis into three groups, which has been subsequent by Boyden. Depending upon the stage of development of the primitive lung bud pulmonary agenesis is classified into three categories : type -1 (agenesis) complete absence of lung and bronchus and no vascular supply to the affected side. type – 2 (agenesis) rudimentary bronchus with complete absence of pulmonary parenchyma.[4]

Type -3 (hypoplasia) presence of variable amount of bronchial tree, pulmonary parenchyma and supporting vasculature diagnosis was suspected from routine radiological examination of the chest where possibilities of atelectasis or agenesis of the left lung was considered. the diagnosis was confirmed by bronchography and pulmonary angiography. The differential diagnosis of this condition in adults include collapse, thickened pleura, destroyed lung and pneumonectomy [5,6] but in some cases angiography is also needed. Surgery is seldom required for agenesis or aplasia as it can be managed on conservative lines. the prognosis in these case depends upon the functional integrity of the remaining lung as well as the presence of associated anomalies. hypoplasia and aplasia are often observed together with malformations (diaphragm defect, kidney anomalies, extra pulmonary sequestration, muscle or skeleton defects). nearly one third of the patients have congenital heart diseases although the most common one is the atrial septal defect, ventricular septal defect, patent ductus arteriosus or coarctation of the aorta can also be observed.

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