Case Report

Unilateral pulmonary agenesis in an adult: A case report from a teaching hospital in Mahabubnagar

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Abstract

Unilateral pulmonary agenesis is a rare congenital bronchopulmonary foregut anomaly; although it is usually diagnosed in infancy and childhood, some cases do not show any symptoms until adult age. We report one case of left sided unilateral pulmonary agenesis in a 45 years old female

Key Words: Chest radiograph, Computed tomography chest (CT), Pulmonary angiogram, Unilateral pulmonary agenesis.

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INTRODUCTION

Unilateral pulmonary agenesis, is a rare congenital bronchopulmonary foregut anomaly, in which there is complete absence of lung tissue on one side. According to Berrocal *et al*., 2004 [1], developmental anomalies of the lung at the 4th and 24th weeks of gestation may the cause bronchopulmonary foregut anomalies. It is usually present in the neonatal period or in early child hood, but some of the cases do not show any symptoms until adult age. Lee EY *et al*., 2011 [2], stated that majority of the patients present with severe respiratory distress (or) repeated pulmonary infections and wheezing, where someone may be completely asymptomatic. Unilateral pulmonary agenesis has an associated with other congenital anomalies involving cardiovascular system, Gastro intestinal tract, Central nervous system, Genito Urinary tract and musculoskeletal anomalies of thoracic cage.

CASE PRESENTATION

A 45 years old female came with complaints of shortness of breath, frequent episodes of cough, easy fatigue since 3 months. On chest radiograph there is radiopaque left hemithorax, crowding of ribs on left side, shift of mediastinum to left side, trachea shifted to right side, elevation of left dome of diaphragm, scoliosis of dorsal spine with convexity towards right side.

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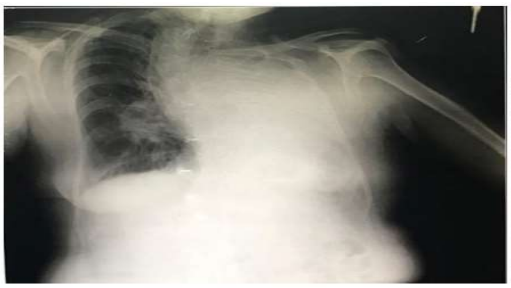
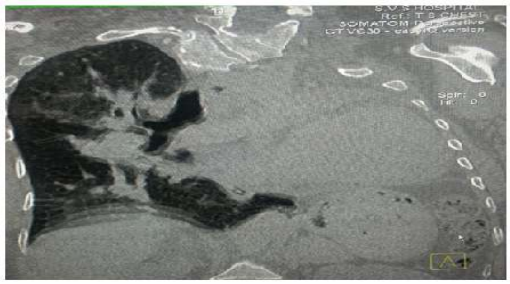


Figure 1



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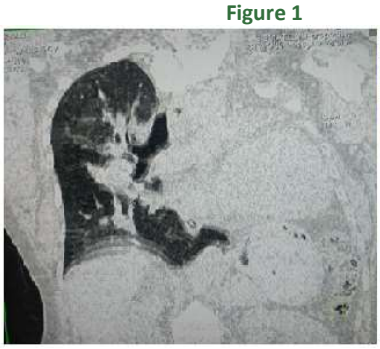

Figure 3

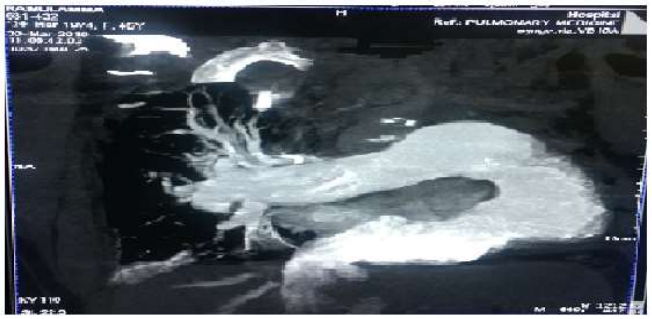

Figure 4

Figure 1: Chest radiograph showing radiopaque left hemithorax, crowding of ribs on left side, shift of mediastinum to left side, trachea shifted to right side, elevation of left dome of diaphragm, scoliosis of dorsal spine with convexity towards right side. On computed tomography chest, reconstructed coronal view at the level of tracheal bifurcation showing absence of left main bronchus; **Figure 2:** CT CHEST – Reconstructed coronal section showing absence of left main bronchus; **Figure 3**: **CT Chest** – Coronal section showing absence of left lung. On pulmonary angiogram, showing only the right pulmonary artery and it's branches, with absence of left pulmonary artery; **Figure 4**: Pulmonary angiogram showing absence of left pulmonary artery.

Provisional diagnosis:

A provisional diagnosis of left lung agenesis was given.

Differential Diagnoses:

Complete left lung atelectasis, left pneumonectomy is ruled out with clinical history and the radiological features.

DISCUSSION

According to Thomas RJ *et al*., 1998 [3], the incidence of unilateral pulmonary agenesis, has been estimated approximately 1 in 15,000 live births or between0.0034% and 0.0097% without sex predilection. Development of the bronchopulmonary tree takes place at about 26th to 31st day of intrauterine life. Many congenital bronchopulmonary malformations characterized by pulmonary developmental anomalies. Schneider *et al*.,1912[4], classified these abnormalities into 3 major types, according to the developmental stage of the primitive pulmonary bud, then modified by boyden as follows.Type 1 (Agenesis): - No trace of lung, bronchus or vascular supply on the affected side, Type 2 (Aplasia): Rudimentory bronchus without any pulmonary tissue and type 3 (Hypoplasia): Presence of variable amounts of bronchial tree, pulmonary parenchyma and supporting vasculature. The present case belongs to the type – 1 of schneider classification. According to Zylak , CJ *et al*., 2002[5], the chest radiographic findings in cases of unilateral pulmonary agenesis characterized by radiopaque ipsilateral hemithorax, contralateral shift of trachea, ipsilateral shift of the mediastinum and crowding of the ribs on affected side. The computed tomography remains the standard modality to establish the degree of under development and to differentiate agenesis from aplasia and hypoplasia, and other conditions, that closely mimic it radiographically. CT shows the absence of tracheal bifurcation and absence of the lung tissue on the affected side [6,7]. CT – pulmonary angiogram is the confirmatory investigation which shows the absence of the ipsilateral pulmonary artery and its branches. The natural history in this case, illustrates the importance of diagnosis of this condition by keeping it in mind as diffential diagnosis in cases, where unilateral homogenous opacity is noted on chest radiograph like, atelectasis and postpneumonectomy[8].

MANAGEMENT

Patient is treated conservatively with oxygen inhalation, antibiotics and other supportive measures.

CONCLUSION

Unilateral pulmonary agenesis is a very rare congenital bronchopulmonary foregut anomaly. Diagnosis of

unilateral pulmonary agenesis requires a high index of clinical suspicious and experience with reading chest radiographs. The radiographic findings will vary depending on the extent of lung involvement and chest CT and pulmonary angiogram are needed to better diagnose this condition. Furthermore, a through congenital anomaly scan during 18 – 22 weeks of pregnancy may help in early diagnosis of pulmonary agenesis and help in better management of this condition.

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