Indian Journal of Medical Case Reports ISSN: 2319–3832(Online)
An Online International Journal Available at http://www.cibtech.org/jcr.htm
2013 Vol.2 (3) July-September, pp.64-69/Kumar et al.

Case Report

A RARE CASE OF AGENESIS OF RIGHT LUNG

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ABSTRACT
Here we are reporting a rare case of agenesis of right lung seen in a male aged 40 years from low socio economic status who was resident of Eastern Uttar Pradesh. He was hospitalized for long standing cough with increased breathlessness. After thorough investigation like CT scan & X-Ray, he was found to have only one lung was present on the left side but absent on the right side. There were no associated conditions and no similar history in the family. Hence this rare anomaly made us interesting to study and report.

Key Words: *Lung Agenesis-Absence of Right Lung - Pulmonary Agenesis - Unilateral Lung Hyporplasia*

INTRODUCTION
Lung agenesis is a congenital anomaly present with respiratory distress. This respiratory distress is due to retention of bronchial secretions, inflammations and poor respiratory reserve. Rarely lung agenesis may be associated with gastric duplication cysts. This gastric duplication of cysts can give rise to nausea, vomiting, hematemesis & pain abdomen (Halilbasic *et al.*, 2013).
Congenital anomalies of lung are very rare anomalies which are sometimes misdiagnosed as tuberculosis. This anomaly if found in teenage, this rare anomaly will be presented in the chest X-Ray as opaque unilateral hemithorax. This anomaly if found in teenage, then this condition of agenesis of lung will be of one of the differential diagnosis. Very often mistaken for unilateral massive pleural effusion with collapse of lung (Bhattacharjee *et al.*, 2012).
Pulmonary agenesis is a rare congenital abnormality of lung (Malcon *et al.*, 2012). Most of them are associated with cardiac & non cardiac anomalies (De, 2013).

CASES
A male aged about 40 years from the eastern part of Uttar Pradesh, farmer by occupation from low socio economic status presented with increased breathlessness with purulent cough for a period of one week.

Past History
He has a very relevant history of hospitalization for similar complaints for nearly 10 times, but he was never investigated because of poverty. Every time, he was given symptomatic treatment in the form of oral salbutamol and theophyllin. He was non-smoker, not a alcoholic. He was son of non consanguineous parents.

On Physical Examination
A male aged 40 years, moderately built and nourished, with no cyanosis, no clubbing and no significant peripheral lymphadenopathy.

On General Examination
Showed features of muscle wasting with loss of lung volume on the right side.
Cardiovascular System revealed normal heart sounds with no murmurs.
On examination of Respiratory System, showed absence of air entry on the right side with few conducted sounds.

Per Abdomen
Soft and no palpable mass felt in any quadrant of the abdomen.
Investigations
a) **Routine blood investigations** were normal.
b) **ECG showed** Right axis deviations.
c) **Pulmonary Function Test (PFT):** Showed Mixed Pattern with Restriction & Obstructions.
d) **Chest X-Ray showed** Complete white out lung on the side.
e) **C-T Scan of Thorax Showed** Complete agenesis of Right Bronchus, Absence of right lung parenchyma, Absence of Right Pulmonary Artery, Absence of Right Pulmonary vein and gross mediastinal shift towards right side.
f) **C-T Scan of Abdomen:** Showed No other congenital anomalies.
g) **2d ECHO**- No Dextro cardia.
Final Diagnosis: Agenesis of right lung.

DISCUSSION
Agenesis of lung is a rare condition in which one of the lungs is absent. There is mediastinal shift along with shift of the heart to the affected side. This anomaly occurs when one of the lung buds fails to develop into full fledged lung (Sudhir, 2008). During the development of lung, there will be division of main bronchus 17 times before birth and 6 times after birth thus forming the bronchial tree. There will be expansion of terminal part of bronchial tree giving rise to alveoli. Mesoderm forms the connective tissue from which arise the pleura. During the intrauterine life, bronchial tree is lined by cubical epithelium. This is the canalicular phase of development of lung. After birth there will be onset of pulmonary circulation where alveoli gets dilated and there will be thinning of lining epithelium (Inderbir and Pal, 2007). A twelve years old boy was diagnosed to have massive pleural effusion with collapse of lung All the relevant investigations like C-T scan of thorax, fibre optic bronchoscopy, along echocardiography were done. Ultimately final diagnosis of unilateral hyporplasia of the lung was made (Bhattacharjee *et al.,* 2012).
A female aged 26 years old had episodic breathlessness tightness in the chest, associated with recurrent nasal obstruction & sneezing due to seasonal variations. Her chest X-Ray was taken which showed opacity of right hemithorax, chest tightness, recurrent nasal obstruction and excessive sneezing, mainly during change of season along with opacity of the right hemithorax in the chest x-ray. A detailed investigation like spirometry, high-resolution CT scan of the thorax and fibre-optic bronchoscopy were done. Her final diagnosis was Right Lung Agenesis (RLA) with bronchial asthma and allergic rhinitis (Kushwaha *et al.,* 2012). A case of pulmonary agenesis was reported in eight months old female child in paediatric department of Medical College Kolkata with history of chronic lung infection due to agenesis of right lung but there were no associated cardiac and noncardiac abnormalities (De, 2013). Two cases of pure agenesis lung were reported from the Pediatrics Department LRS Institute of TB and Respiratory Diseases, New Delhi. In one case in a thirteen years old girl who came history of infection of the chest for the first time, not responding to medical line of treatment was found to have isolated agenesis of the lung and other also a female child ten years old had history of illness of chest infection along with pulmonary tuberculosis for a period of one year & at the end one year she found to have agenesis of upper lobe of left lung & in addition positive report of Mycobacterium Tuberculosis in gastric lavage (aspirate). Later she was put antituberculous line of treatment and she responded well (Sharma *et al.,* 2005).
Present Study
In the present study, a rare case of agenesis of right lung has been studied. This study was done in a male from eastern part of Uttar Pradesh who came to outpatient department with a history of persistent productive cough associated with breathlessness since 5 days. He was thoroughly investigated and found to have absence of right lung along with absence of other vessels on the right side. He was not smoker,

not an alcoholic not a tuberculosis patient. He was not diabetic, not a hypertensive & not born to consanguineous parents. He was diagnosed to have absence of right lung there is compensatory enlargement of left lung. There were no associated conditions like bronchial asthma and allergic rhinitis as seen in studies of Kushwaha *et al.*, (2012) nor a gastric duplication cyst as seen in studies of Halilbasic *et al.*, (2013). There were no cases of tuberculosis as seen in studies of Sharma *et al.*, (2005). So our study is purely of agenesis of right lung which made us interesting to study this case and report.

Take Home Message:

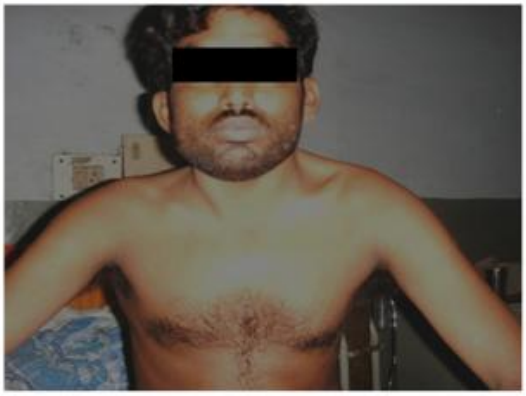

Figure 1: Showing drooping of right shoulder and pectoral muscle right side in male aged 40 year

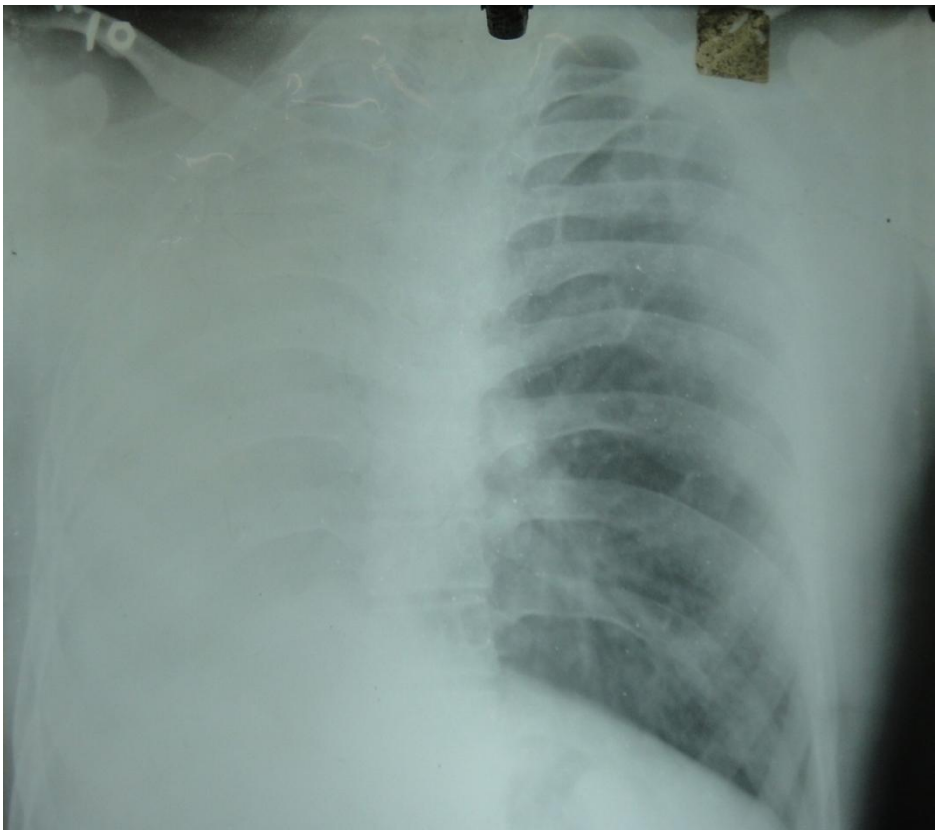

Figure 2: Complete soft tissue homogenous opacification of right lung. Mild rib crowding noted on right side

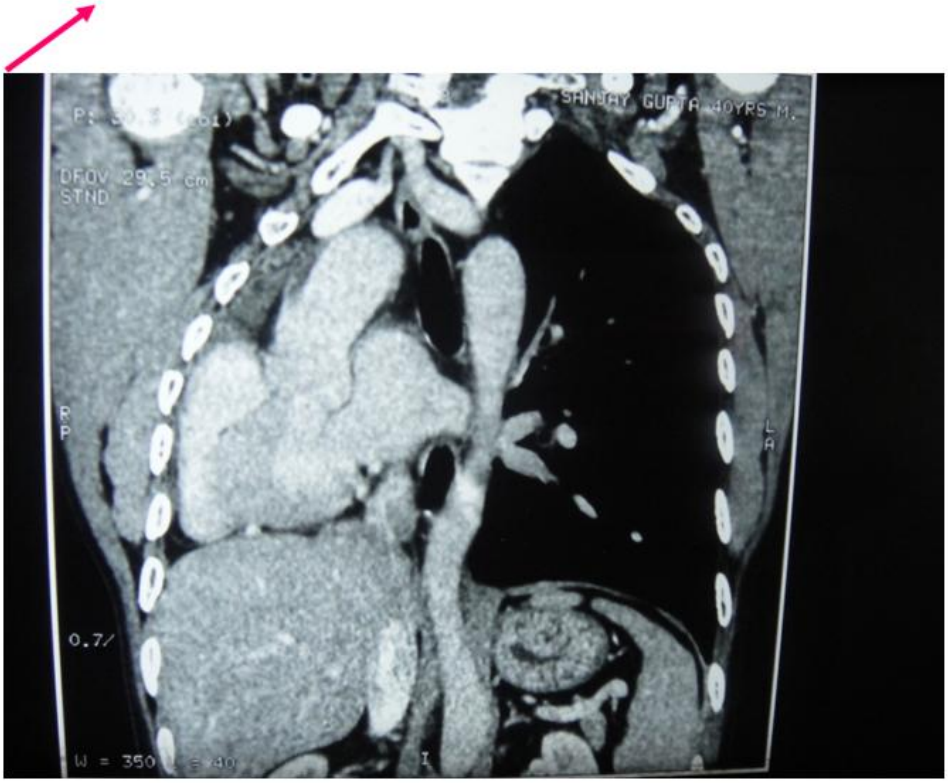

Figure 3: CT coronal section of thorax- showing trachea, aorta, Heart, left pulmonary artery and gross mediastinal shift towards right side (no dextro cardia)

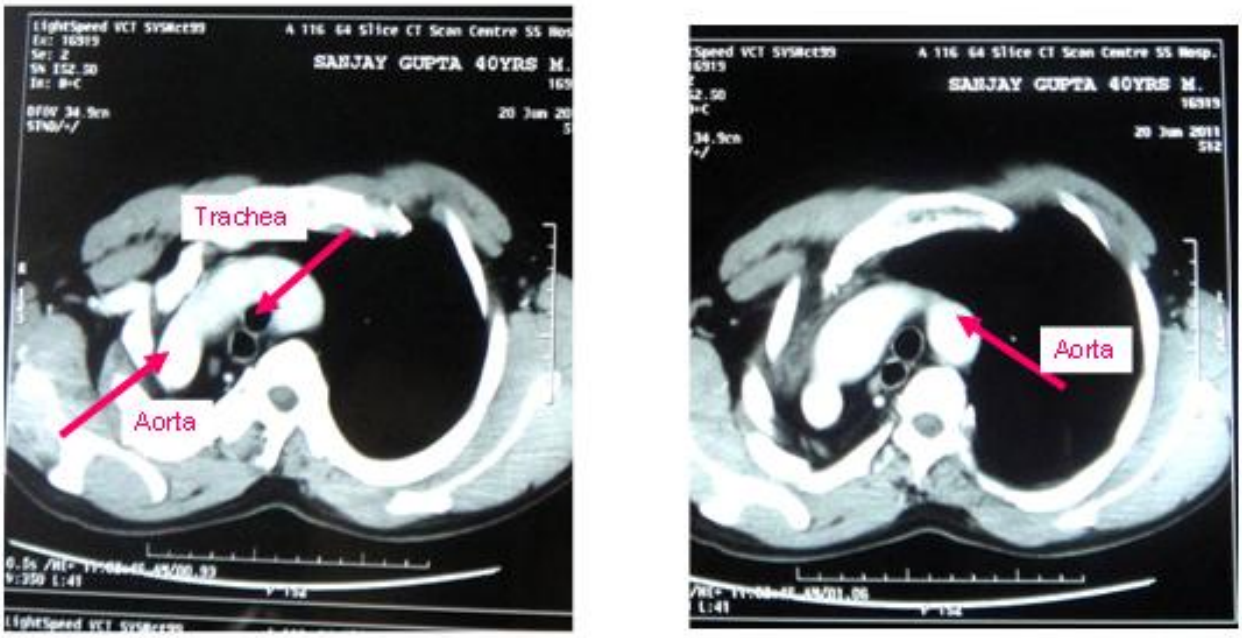

Figure 4: trachea, aorta and mediastinal shift towards right side

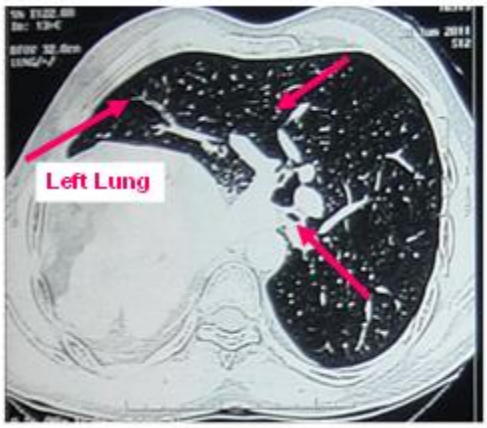

Figure 5: Absence of right bronchus, right side lung parenchyma, right pulmonary artery and right pulmonary vein

a) **Complete absence of right bronchus**
b) **Marked mediastinal shift and cardiac rotation with proliferation of fat into the right thorax**
c) **Compensatory hyper ventilation of anterior herniation of the left lung**
d) **Right pulmonary artery and veins are not visualized with main pulmonary artery is seen continuing on left side**

Since it is case of congenital anomaly of lung commonly seen in paediatric age group, but rarely seen in Adult age groups. This has profound clinical importance in thoracic medicine. They are difficult to diagnose because they come with infection of lung with breathlessness & productive cough mistaking it to be infection of lung but with proper clinical history supported by relevant investigations agenesis of lung has to be diagnosed Hence studied this rare entity.

ACKNOWLEDGEMENT

We sincerely thank

1) Forty years old from UP who allowed us to examine on him and to carry out relevant investigations.
2) Chief Editor of publishers for accepting our case report for reporting our case of Agenesis of Right Lung.

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