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Right Lung Aplasia with bronchial asthma , a rare case

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INTRODUCTION: Pulmonary aplasia is a rare congenital anomaly with reported prevalence of 34 per ten lakh live births. Arrested development of primitive lung bud leads to anomalies of agenesis, aplasia and hypoplasia. Right lung agenesis/aplasia has dismal prognosis compared to left side due to frequent association with a gamut of other congenital anomalies and greater degree of mediastinal shift leading to tracheobronchial and vascular distortion.

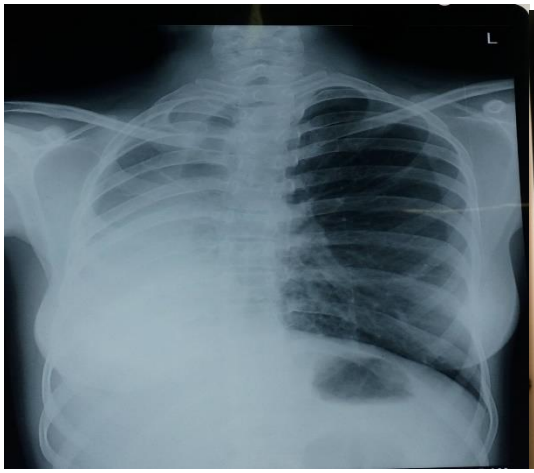
CASE REPORT: A 25 year old female patient presented with complaints of cough with expectoration and breathlessness since 2 months . History of frequent wheezing was present and no history of fever, hemoptysis , chest pain . History of similar complaints were present since 10 years . On examination, she was thinly built, moderately nourished. Trachea and apex beat were markedly shifted to right side .Right side of chest was slightly retracted, vocal fremitus was decreased and there was dull note on percussion. . Breath sounds were absent over most of right lung field except in the infra clavicular area where vesicular breathing was heard. Expiratory polyphonic rhonchi were heard over left lung .

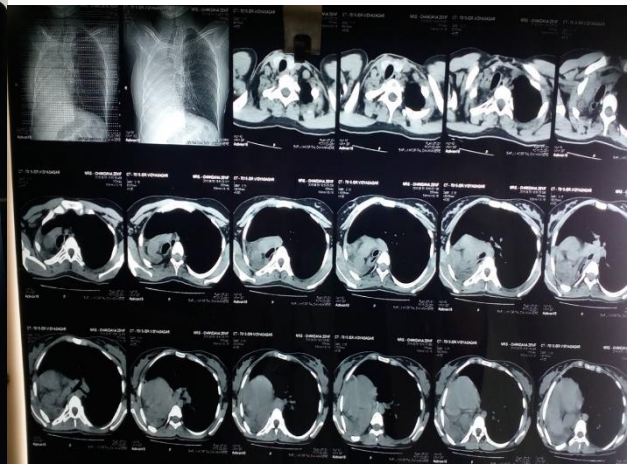
Examination of other systems was found to be normal .

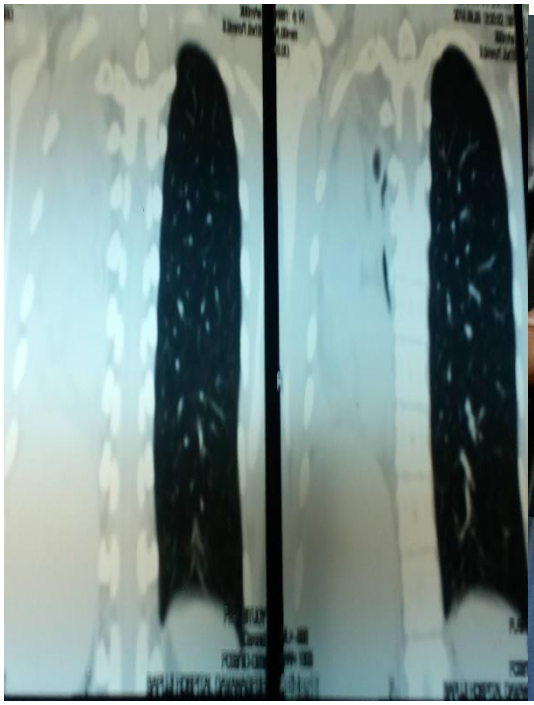
Chest x ray and HRCT showed right homogenous opacity with ipsilateral mediastinal shift.

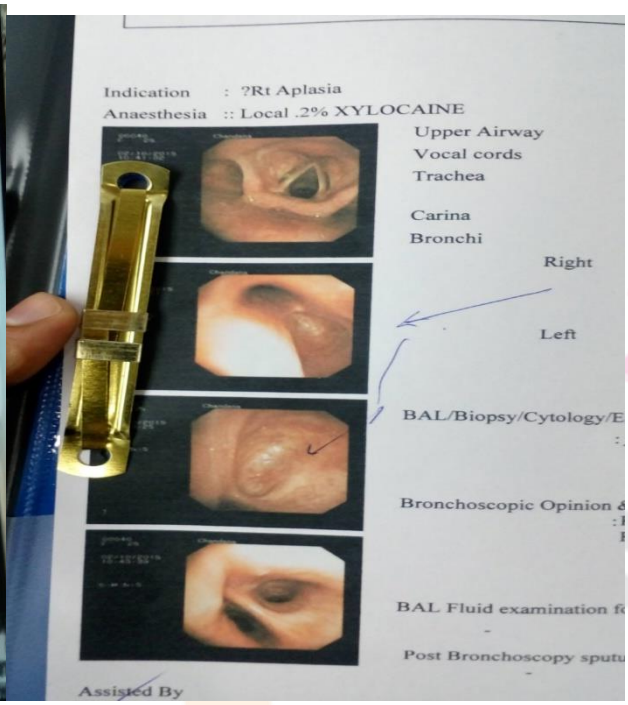
On flexible bronchoscopy, carina was widened, mucosa was healthy, there was no scarring and right main bronchus ended abruptly. The scope could enter the opening of this 1 cm pouch. 10 ml of normal saline was instilled into the pouch and was almost completely aspirated.

USG abdomen showed normal study.









PULMONARY FUNCTION TEST

	Predicted (L)	PRE (L)	%PRED	POST(L)	%PRED	%Change
FVC	2.56	1.78	69.5%	1.67	65.3	-6.0
FEV1	2.38	1.09	45.7	1.15	48.2	5.4
FEV1/ FVC (%)	84.53	61.01	72.3	68.40	81.1	7.4
FEF 25-75 (L/ S)	3.98	0.89	22.2	0.94	23.7	6.7

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MANAGEMENT; Advised long acting beta agonist with Inhalational corticosteroids .She improved with inhalational medication .Regular follow up was advised.

DISCUSSION Agenesis of the lung is an extremely rare congenital anomaly representing failure development of the primitive lung bud. This condition was first described by De POZZE[1].Schneider[2] in 1912 classified agenesis into three groups, which have

been modified by Boyden. Depending upon the stage of development of the primitive lung bud, pulmonary agenesis is classified into three categories: **Agenesis** - Complete absence of lung and bronchus and no vascular supply to the affected side.

Aplasia - Rudimentary bronchus with complete absence of pulmonary parenchyma.
Hypoplasia - Presence of variable amounts of bronchial tree, pulmonary parenchyma and supporting vasculature.
Nearly 50% cases of pulmonary agenesis have associated congenital defects[4], which usually involve cardiovascular, skeletal, gastrointestinal and genitourinary system. The exact etiology of this condition is unknown though genetic factors, viral agents and dietary deficiency of Vitamin A during pregnancy have been implicated[4]. Left sided agenesis is more common and the subjects have a longer life expectancy than those with right sided agenesis[3]. This is probably due to excessive mediastinal shift and malrotation of carina in right sided agenesis which hinders proper drainage of the functioning lung and increases chances of respiratory infections .Final diagnosis can only be established after bronchoscopy and in some cases angiography. Surgery is seldom required for agenesis or aplasia, which can be managed on conservative lines. The prognosis in such cases depends upon the functional integrity of the remaining lung as well as upon the presence of associated anomalies

Our patient presented with recurrent respiratory complaints and diagnosis was suspected from routine radiological examination of chest, where possibilities of atelectasis or aplasia of the right lung were considered. The diagnosis was confirmed by bronchoscopy and CT chest. 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. However, patients with one lung have been reported to survive well into adulthood without much complaints

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