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A RARE CASE OF VANISHING LUNG SYNDROME WITH HEPATITIS-B

Dr. Pulkit Gupta[1]*, Dr. G. N. Srivastava[2], Dr. Anbarasan Thiru[a], Dr. A. Tiwari[1], Dr. A. Jain[a], Dr. S. S. Dash[1], Dr. A. Suresh[1], Dr. Gowthami B.[1] and Dr. K. S. Sravani[1]

[1]Junior Resident, Dept. of TB & Respiratory Diseases, IMS, BHU.
[2]Prof., Dept. of TB & Respiratory Diseases, IMS, BHU.

***Corresponding Author: Dr. Pulkit Gupta**
Junior Resident, Dept. of TB & Respiratory Diseases, IMS, BHU.

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ABSTRACT
Vanishing lung syndrome (VLS) is a rare disorder otherwise known as idiopathic giant bullous emphysema, typically occurs in young, thin male smokers but cases have been diagnosed in middle aged men as well. In this disorder emphysematous bullae appear in the lung, alveolar walls gradually disintegrate to form large air spaces with eventual atrophy of the lung and[1-3] and compression of the adjacent side. The basic pathogenesis is not exactly known, however it has been attributed to[2] possible respiratory myositis. Vanishing Lung Syndrome was first described in a case report by Burke in 1937.[4] Since then several more case reports have been published in various journals worldwide.[5-8] It is most often confused with pneumothorax and inadvertent insertion of intercostal drainage is not a rarity even among reported cases.[5,8] The patients have little or no evidence of cough or respiratory infection[1] and finally die in respiratory failure. The radiographic criteria for this syndrome defined by Roberts et al include the presence of giant bullae in one or both upper lobes, occupying at least one third of the hemithorax and[9] compressing surrounding normal parenchyma. VLS is also known as type I bullous disease or primary bullous disease of the lung in which the lungs appear to be[10] disappearing on X-ray. Surgical resection of giant bullae is the treatment of choice, the indications include spontaneous pneumothorax, infection, or dyspnoea, the[11-15] best results are seen following limited bullectomy. Thoracoscopic treatment of giant bullae is an effective alternative to conventional thoracotomy with minimal[16] morbidity.

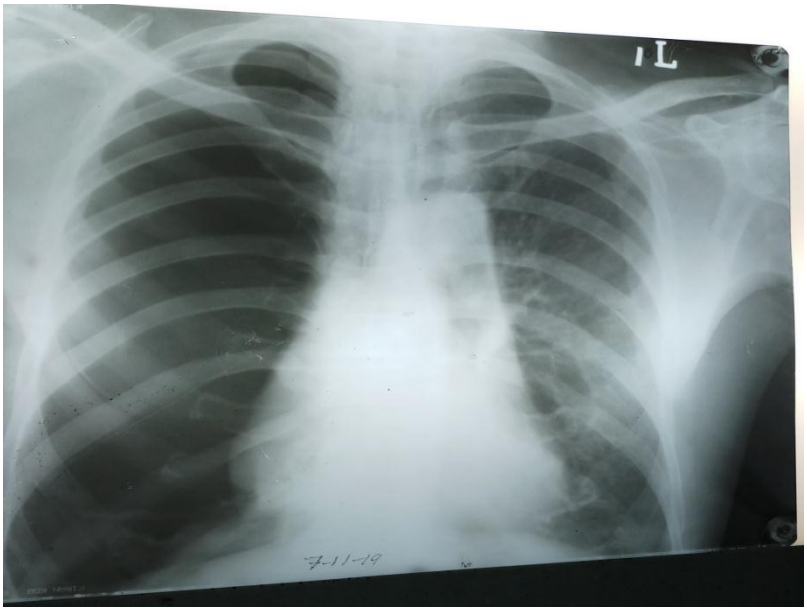
KEYWORDS: spontaneous pneumothorax, infection, or dyspnea.

CASE REPORT
A 55 years old male, farmer by occupation presented to us with chief complaints of shortness of breath since two years (increased since two months), cough and right sided chest pain since two months. Shortness of breath was insidious in onset, mMRC grade 2 initially but grade 3 since 2 months. It aggravated on exertion and was relieved with rest. Patient also complained of cough since two months. It was dry in nature and was not associated with diurnal, seasonal or postural variation.He had chest pain for two months, which was insidious in onset and dull aching in nature. It was localized to right hemi-thorax and was non-radiating and non-migratory. Itwas aggravated on exertion and relieved with rest.

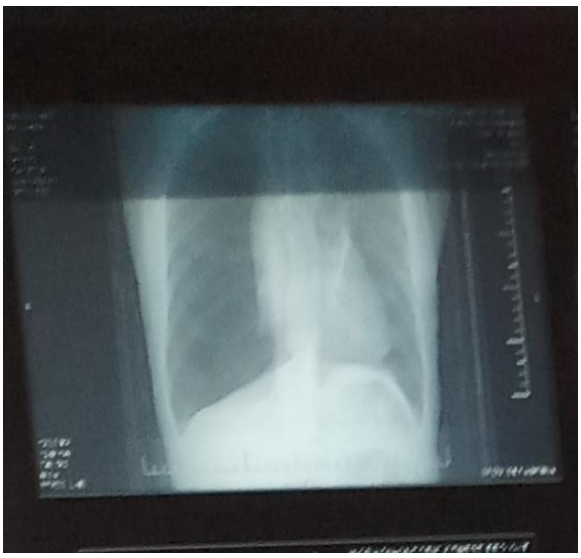
No complaints of fever, hemoptysis, loss of weight, loss of appetite, orthopnea, abdominal pain, nausea, vomiting, trauma, altered sensorium or loss of consciousness.

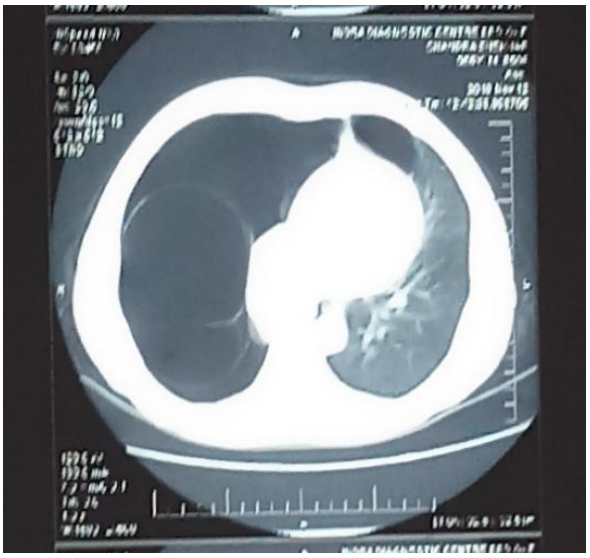
No history of tuberculosis / DM / HTN / CAD / thyroid disorders / asthma or inhaler use in the past. Smoking history- patient is a smoker with smoking history of cigarettes and marijuahna since 25 years. On general examination there was no pallor, icterus, clubbing, cyanosis, pedal edema or lymphadenopathy. JVP was not raised. Then respiratory system was examined. On inspection, there was asymmetrical chest movement with decreased movements on right side. On palpation, chest movement decreased on right side and trachea shifted towards left, vocal fremitus decreased on right side in all regions. On percussion, hyper-resonant notes were present in all regions of right hemithorax and upon auscultation air entry was absent on right side.

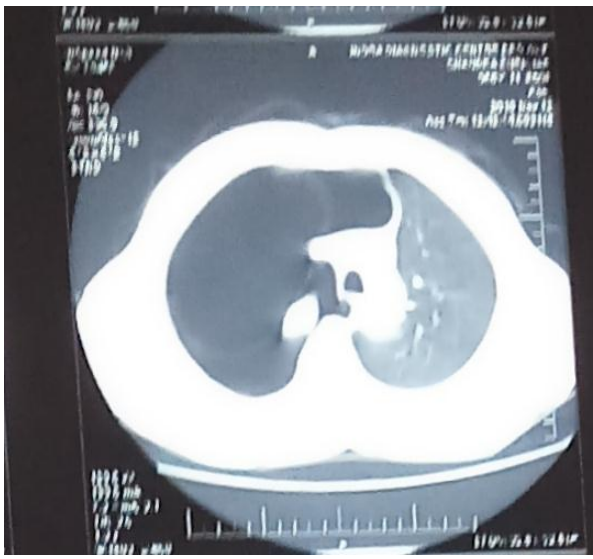
Investigations
X-ray chest- patient had an old x-ray at the time of presentation. It showed the presence of right sided hyperlucency.

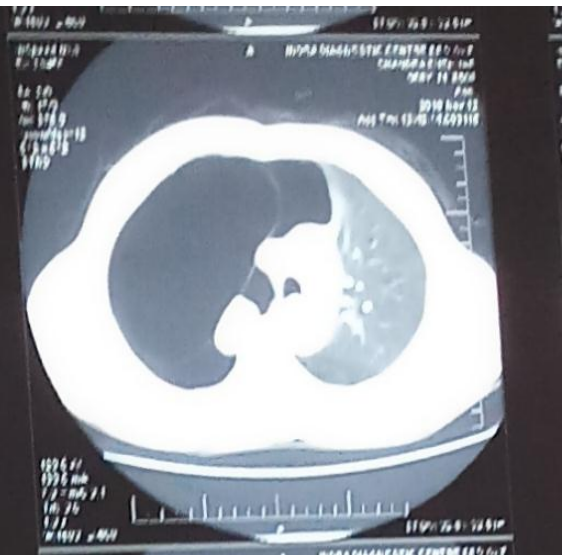


Suspecting it to be a case of bullous lung disease, then CECT thorax was done.









Findings of CECT thorax- increased translucency in right lung field seen.
-gaint emphysematous bullae occupying almost the whole right lung.
-normal vascular markings are present in left lung.
-widening of intercostal space on right side seen.

Laboratory parameters like ABG, CBC, LFT, RFT, RBS, ECG, Cardiac profile, d-dimer was normal including α-1 antitrypsin (151, normal = 120-200).
Hepatitis B Surface Antigen was positive.
PFT- restrictive pattern was obtained

DIAGNOSIS

As there was a giant bullae in hemithorax(right side), occupying more than one third of the hemithorax, it was diagnosed as a case of Vanishing Lung Syndrome. Patient also had hepatitis-B infection which was newly diagnosed.

TREATMENT AND FOLLOW-UP: Treatment of choice is surgical resection of giant bullae. Bullectomy is performed based on eligibility criteria which includes smoking cessation, exercise capacity, pulmonary function tests (FEV_1>35%) and age less than 50 years.

Our patient did not meet the criteria for surgical intervention. So the patient was discharged with conservative management and proper counselling about smoking cessation. Patient is also receiving hepatitis-B treatment. Patient is on regular follow up with us and is doing well.

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