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Case Report

Internal Medicine Section

A Case of Intralobar Pulmonary Sequestration Diagnosed Incidentally in a Geriatric Male

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ABSTRACT

Pulmonary sequestration is a rare congenital malformation of lung in which a part of the lung is supplied by an anomalous systemic blood supply from thoracic or abdominal aorta. This is a nonfunctioning lung tissue and lacks normal communication with tracheobronchial tree. It develops from accessory lung bud. If developed before pleura formation, it results in intralobar sequestration, and extralobar, if it develops after pleura formation. Usually Intralobar type of pulmonary sequestration presents in late childhood or adolescence with recurrent pulmonary infections. It is less commonly associated with foregut communication or associated anomalies, unlike the latter. Extra lobar type usually presents with respiratory distress while recurrent infections are less common. In the present case report a 76-year-old male patient was diagnosed with intralobar pulmonary sequestration. This patient was presented with recurrent respiratory infections with complaints of fever and cough with expectoration for one week. Previously the patient was misdiagnosed with recurrent pneumonia. Despite the age of patient, consideration of pulmonary sequestration as a differential diagnosis lead to investigations in that direction. This helped in prompt diagnosis and further plan of management. Misdiagnosis of this condition may have lead to development of complications like recurrent infections and abscess. These complications can be avoided by prompt surgical excisions which are curative. This case report was unique because it diagnosed pulmonary sequestration in a 76 years old patient who presented with recurrent pneumonia.

Keywords: Bronchopulmonary sequestration, Computed tomography angiography, Congenital anomaly, Recurrent pneumonia, Video assisted thoracic surgery

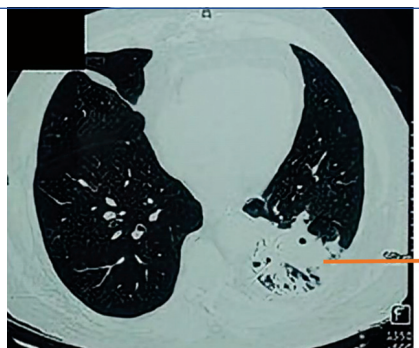
CASE REPORT

A 76-year-old male, with history of frequent lower respiratory tract infection came to the Outpatient Department (OPD) of Department of Respiratory Medicine with complaints of fever and cough with expectoration for one week. At the time of presentation, the patient also had left-sided chest discomfort since one week and breathlessness Grade II Modified Medical Research Council (MMRC) [1] since one week.

The past history of the patient showed visits to local doctor for symptomatic relief from seasonal flu and respiratory symptoms being diagnosed as pneumonia. There was no history of any co-morbidity and major medical or surgical illness. The patient was a tobacco chewer amounting to one to two packets of tobacco/day for 30 years. Family history was non-significant.

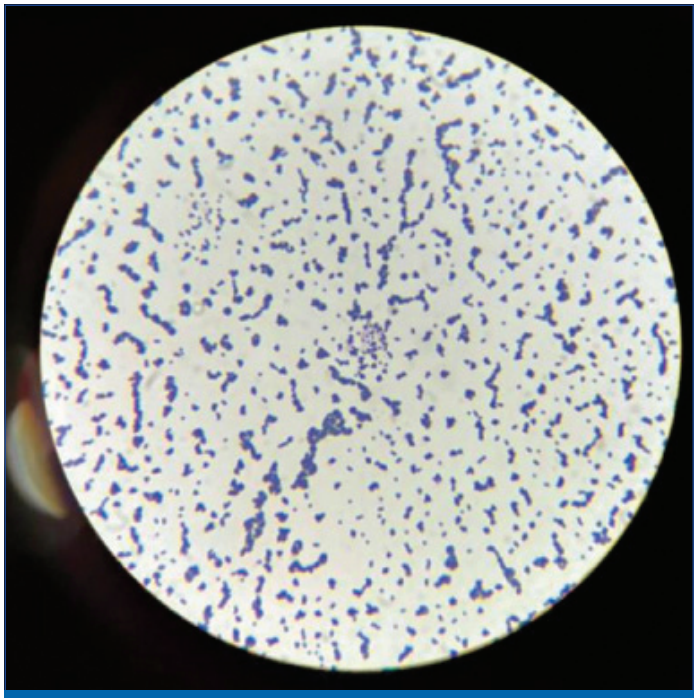
Physical examination suggested decreased respiratory movements in left mammary, inframammary and infra-axillary areas. The patients vitals were stable and within normal limits. On auscultation of chest, coarse crepitations were audible in left basal area, left inframammary, infra-axillary, infrascapular areas with decreased breath sounds in same areas. Laboratory results showed moderate leukocytosis. Other blood parameters like random blood sugar, serum electrolytes, liver and kidney function tests were within normal limits.

Computed Tomography (CT) scan revealed Intralobar, lobulated lesion of approximately 2.5×3.5×4 cm in size with irregular margins in the postero basal segment of left lower lobe having a heterogeneous appearance with central cystic spaces with areas of ground glass opacities and consolidation [Table/Fig-1]. A 3 Dimensional (3D)-Multiplanar Reconstructions (MPR) images showed that the lesion was supplied by a feeding artery originating from the descending thoracic aorta coursing superolaterally and drained by vein into inferior pulmonary vein [Table/Fig-2]. The aberrant artery measured 5-7 mm in diameter. On basis of the clinical findings alone differentials suspected were pneumonia, tuberculosis, bronchiectasis and abscess. Radiological findings from CT and 3D-MPR showed blood supply of the lesion lead to consideration of diagnosis of Intralobar Broncho pulmonary Sequestration (BPS). Induced sputum samples of the patient were sent for investigation considering secondary bacterial infection, fungal infection or tuberculosis. They revealed Gram positive cocci [Table/Fig-3]. Acid fast bacilli or fungal pathogen were not seen. The patient was also advised to undergo bronchoscopy to rule out local bronchial airway obstruction, collect Bronchoalveolar Lavage (BAL) sample to rule out tuberculosis and also to identify the organism in case of non resolving pneumonia. The final diagnosis of intralobar pulmonary sequestration was made and patient was referred to Cardio-Thoracic Surgical Department. The patient was given surgical option of feeding artery embolization, Video Assisted Thoracic Surgery (VATS) and lobectomy but patient denied for any type of invasive treatment due to personal reasons and was thus discharged against medical advice after being stabilised on symptomatic treatment which included antipyretics, cough expectorants, mucolytic, antacids and broad-spectrum antibiotic (combination of cifixime and clavulenic acid). The patient was lost to follow-up and was untraceable even after multiple attempts to contact.



[Table/Fig-1]: CT thorax shows posterobasal segments of left lower lobe with heterogeneous appearance and central cystic spaces with areas of ground glass opacities with consolidation.

[Table/Fig-2]: 3D-MPR images showing left postero basal lesion is supplied by a feeding artery originating from the descending thoracic aorta coursing supero-laterally and drained by vein into inferior pulmonary vein.



[Table/Fig-3]: Sputum examination showing gram positive cocci. (10x magnification, Gram stain)

DISCUSSION

Broncho pulmonary sequestration is a rare congenital malformation in which part of the lung derives its blood supply from an anomalous systemic artery [2]. It comprises of about 0.15-6.4% of all congenital pulmonary malformations and usually the posterior basal segment of the lung is commonly affected [3]. Gender wise incidence shows 51% males and 49% females [4].

One of the most widely accepted hypothesis suggests that it occurs from development of an accessory lung bud which develops inferior to the normal lung buds during embryogenesis. Also, this "accessory" lung bud gets its individual aberrant feeding vascular supply which is mostly from the aorta [5]. Many times it is diagnosed during sonography follow-ups in antenatal clinic visits [6].

Broncho pulmonary sequestration can be intralobar or extra lobar. Intralobar sequestration is most commonly associated with infectious complications like recurrent pneumonias. On radiological evaluation of recurrent infections, broncho pulmonary sequestration is often an incidental finding [7,8].

In this case study, the pulmonary lesion was on left posterobasal side and intralobar type. Although in a case report by Phelps MC et al., about 70 % of intralobar pulmonary sequestrations were found to be in left lower lobe and about 30% cases were located in right lower lobe. The BPS was found in the right lower lobe [8]. Likewise, in a case report by Cooke CR, right lower lobe was the site affected in a case of BPS [5].

CT Angiography (CTA) has been found to be the non invasive investigation of choice for diagnosing BPS In a retrospective analysis of 43 cases done by Long Q et al. Multidetector CT successfully detected BPS in all 43 patients (100%) along with related parenchymal characteristics, arterial supply and venous drainage which also helped to plan surgical strategies [9]. Lin CH et al. did retrospective review of 31 patients who underwent surgical procedures for BPS, of which 30 patients were diagnosed by CTA. The CTA was concluded to be the non invasive procedure of choice for diagnosis and evaluation of vascular supply of BPS This also helped preoperative evaluation of aberrant vessels to avoid complications [10].

The typical presentation of this condition is manifested by the second decade of life. Its diagnosis can be missed in the early life due to overlapping of many respiratory symptoms and radiological findings of other respiratory conditions [11]. Many patients present in geriatric age group after development of common complication of pneumonia or other complications like malignancies, pneumothorax, haemoptysis or haemothorax [12]. But in this case scenario, the patient was presented with mild chest discomfort, breathlessness along with symptoms of cough with expectoration and fever. So in a patient presenting with recurrent pulmonary infection, possibility of pulmonary sequestration should be considered. As with a high index of suspicion, misdiagnosis of patient can be avoided. Other differential pulmonary conditions associated with BPS along with their radiological appearance are Infected cystic bronchiectasis (saccular dilation of bronchi on imaging), bronchogenic cyst (well circumscribed spherical or ovoid masses on imaging), congenital diaphragmatic hernia (multiple air fluid levels on chest imaging, scaphoid abdomen and gurgling sound in chest on clinical examination), cystic adenomatoid malformation (multiple air fluid levels on chest imaging, usually diagnosed antenataly) [13]. The differential diagnoses to evaluate before establishing definitive diagnosis of BPS include, lower respiratory tract infections leading to pneumonia, fungal infection, bronchiectasis, abscess. Induced sputum examination can be used to rule these out. Other diagnoses considered were congenital malformations like bronchogenic cysts, mediastinal cysts (mediastinal widening on chest imaging), diaphragmatic hernia, cystic adenomatoid malformation, non malignant and malignant mediastinal or paravertebral mass (mediastinal widening and/or lymph node involvement on chest imaging, features of dysphagia and cough on clinical examination) [14,15]. Each of these diagnoses were ruled out after confirmation of absence of their particular characteristics on CT imaging.

In patients with recurrent symptoms, the most definitive management available is surgery. Surgical intervention comprises of lobar resection by either standard thoracotomy or VATS and endovascular embolization of its feeding systemic vessel. For asymptomatic patients and not consenting for surgery, regular observation should be done for signs of any pulmonary complication. While in patients with recurrent respiratory symptoms, surgery is the most definite treatment [16]. However the decision regarding surgical resection is often weighted against various factors including clinical manifestations, risk of surgical complications, co-morbidities, and patient preferences [17]. In this case scenario, patient was advised interventional and surgical management to which he refused. So he was treated symptomatically and was lost to follow-up.

CONCLUSION(S)

In present case report, diagnosis of intralobar pulmonary sequestration had been made in patient with history of frequent lower respiratory tract infections.Proper radiological evaluation using CT and 3-D MPR led to prompt diagnosis. Broncho pulmonary sequestration is a congenital manifestation. But it should

always be considered in cases of recurrent lower respiratory tract infections and recurrent pneumonias in adults as in present case. This enhances the chance of a parenchymal-sparing resection if surgical intervention is considered. The decision regarding surgical resection is to be weighed against various factors including clinical manifestations, risk of surgical complications, co-morbidities, and individual patient preferences.

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