

Bronchopulmonary sequestration: A rare congenital anomaly presented in an adult female patient

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Abstract

Pulmonary sequestration is a congenital anomaly of lung parenchyma without a normal connection to the tracheobronchial tree and an anomalous systemic arterial supply. The presentation of this condition is variable ranging from no symptoms to hemoptysis. We report a case of pulmonary sequestration in an adult female who presented with recurrent chest infections. Her chest x-ray showed non-homogenous opacity in left lower lobe of lung and CT scan findings were suggestive of intralobar pulmonary sequestration in left lower lobe having arterial blood supply from the descending aorta and venous drainage in left inferior pulmonary vein. She subsequently underwent resection of the left, lower lobe of the lung.

Introduction

Bronchopulmonary sequestration represents a spectrum of abnormalities [1]. Pulmonary sequestration refers to the situation whereby a portion of lung tissue receives its blood supply from an anomalous systemic artery [2]. Two types of pulmonary sequestration are recognized [3-10], depending on whether or not the malformation possesses its own pleural covering. Intralobar sequestration is an abnormal region within the normal pulmonary parenchyma without its own pleural covering. Extralobar sequestration corresponds to a true accessory lung, with its own pleural envelope. Intralobar sequestration accounts for 0.15% to 1.7% of all congenital lung abnormalities [11]. Numerous reports have described serious complications arising from both intralobar as well as extralobar pulmonary sequestration such as fungal infections, tuberculosis, fatal hemoptysis, massive hemothorax, cardiovascular problems, benign tumors and even malignant degeneration [12]. We report this case to increase awareness about the condition and to highlight the significance of considering this rare etiology in a differential diagnosis of adult patients who present with recurrent pulmonary infections.

Case report

27 years old female presented in OPD with complaints of left sided chest pain, productive cough off and on for 2 years. She had a history of recurrent respiratory tract infections. She was previously diagnosed as having tuberculosis and had taken 2 courses of Antituberculous therapy, 13, and 11 years back, with no resolution of symptoms. On examination patient had

mild anemia with dull percussion note and decreased air entry on left side of lower chest. The chest radiograph showed non homogenous opacity in the left lower zone of chest. CT scan showed multiple cystic spaces in left lower lobe with thin enhancing septae. Air specks were seen suggestive of infection. Enlarged paraesophageal, left hilar and perivascular lymph nodes were also seen. Findings were suggestive of intralobar pulmonary sequestration in left lower lobe having arterial blood supply from the descending aorta behind the left crus of diaphragm and venous drainage in left inferior pulmonary

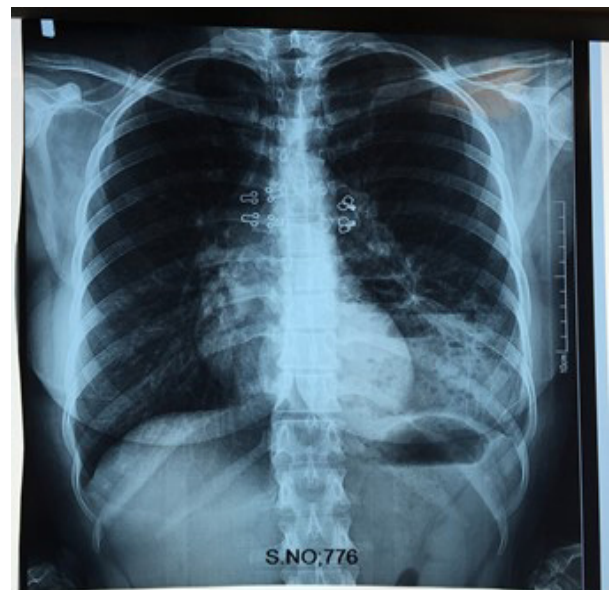


Figure 1: Chest x- ray showing non homogenous opacity in left lower zone.

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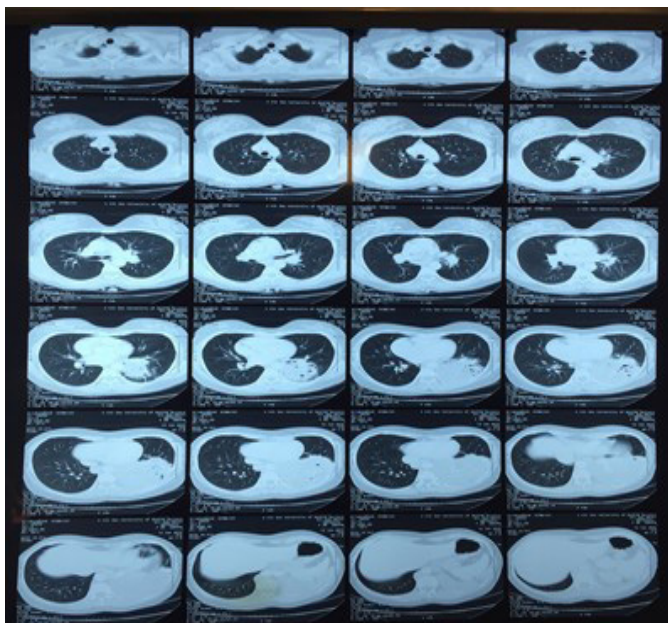


Figure 2: CT-scan chest showing non homogenous opacity in left lobe of lung.

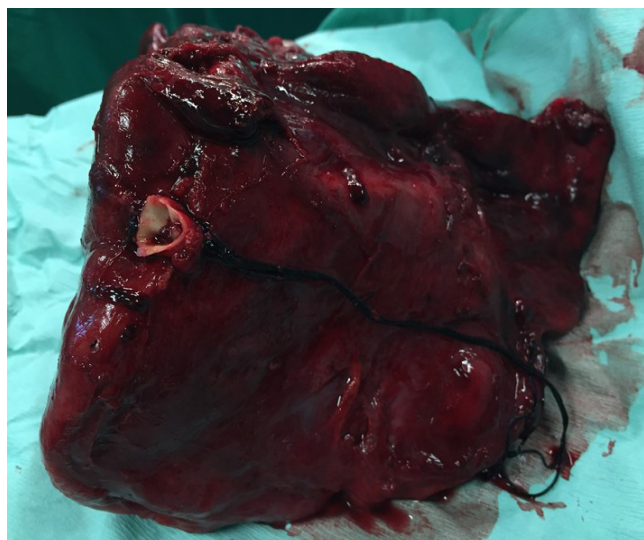


Figure 4: Showing resected left lower lobe of lung.

vein. No definite communication with tracheobronchial tree was seen. Overall appearance was suggestive of intralobar pulmonary sequestration with superimposed infection. A diagnosis of Left lower lobe type 3 pulmonary sequestration with bronchiectasis was made.

After pre-operative workup and anesthesia fitness, left thoracotomy and left lower lobectomy was performed. Per-operatively thick and discolored lower lobe of left lung with pus filled cystic spaces were found. The aberrant systemic artery of approximately 1cm caliber arising directly from aorta entering the posterior segment of lower lobe of the left lung was ligated. Post operatively patient remained stable, and the chest roentgenogram was within normal limits and patient was discharged on 10th postoperative day.



Figure 3: CT-scan showing multiple cystic spaces in left lower lobe.

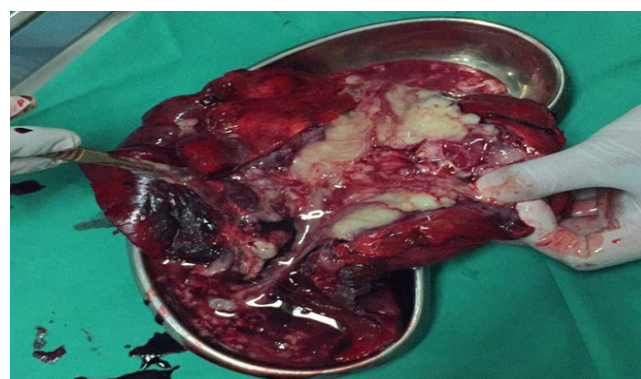


Figure 5: Showing sequestered tissue inside the left lower lobe of lung.

Discussion

Pulmonary sequestration is a relatively rare entity comprising 0.15%-0.4% of all congenital malformations.¹³ It occurs when a disturbance in embryonic development produces a cystic mass of non-functioning lung tissue. Most often the mass is supplied by an anomalous artery and has its own bronchial system, which usually does not communicate with the normal bronchial tree [14]. Pryce for the first time described this as a condition in which an abnormal artery is associated with an ectopic pulmonary mass in the lower lobe of lung [15]. Sequestrations are classified basically as, intralobar and extralobar. Intralobar according to the Pryce is classified into 3 types. Type 1 consists of regularly ventilated lung tissue perfused by two arterial blood supplies on the margins (pulmonary artery, systemic artery), type 2 of sequestered, irregularly ventilated (atelectatic) lung tissue perfused by two arterial blood supplies on the margins (pulmonary artery, systemic artery) and type 3 of sequestered, not ventilated lung tissue perfused only by the systemic artery blood supply. Intralobar sequestration usually presents in adulthood and most commonly involves of left lower lobe [16,17]. Occasionally it is an incidental finding on roentgenograms of the chest in an asymptomatic patient; however the disorder is usually symptomatic and the most common presentation is recurrent pulmonary infections [18]. Plain chest radiograph is usually

nonspecific, showing an ill-defined consolidation that mimics pneumonia, or shows a solitary soft tissue mass or nodule, or a cystic or multicystic lesion [19,20]. Chest CT usually shows a discrete mass in the posterior- or medial-basal segment of the lower lobe, with (as in our case) or without cystic changes [21]. Our patient also presented in a way intralobar pulmonary sequestration commonly presents. The standard treatment is resection of the segment or lobe that contains the sequestered tissue; the prognosis is favorable [22,23].

Conclusion

Bronchopulmonary sequestration can present with multiple nonspecific findings. This case illustrates a typical presentation of an intralobar bronchopulmonary sequestration and highlights the importance of considering this rare congenital condition when treating recurrent chest infection in young adults so that timely identification and management could be undertaken.

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