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Case Report

Pulmonary sequestration: An uncommon presentation with acute chest pain

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ABSTRACT

This is a case of 18-year-old teenager presented with acute left sided chest pain for five days. This was associated with intermittent cough, and loss of weight in two weeks. Diagnosis was made by computed tomography of thorax plus angiogram that showed infected left intralobar pulmonary sequestration with lung abscess. Sputum culture grew *Pseudomonas aeruginosa*. He completed 14 days of antibiotic and subjected to feeding artery embolization. The aim of this case report is to highlight on the uncommon presentation and the need of high index of suspicion of pulmonary sequestration with support by imaging findings.

1. Introduction

Bronchopulmonary sequestration or pulmonary sequestration (PS), is a rare congenital pulmonary malformation that has an incidence between 0.15% and 6% [1,2]. It is a non-functional lung tissues or lobe with absence of communication with bronchopulmonary tree. It has its own anomalous vascular supply. PS can be divided into two types; intralobar and extralobar, with intralobar more commonly found [3]. Patient can be asymptomatic or presents with symptoms, commonly of recurrent lung infection and less commonly, chest pain and hemoptysis that could mimics other diagnoses.

Chest pain in young adult is not common and when it occurs, it should raise a suspicion of underlying sinister pathology or disease. In PS, chest pain is not the most reported clinical presentation. This case helps to highlight the consideration of PS as a diagnosis in young adult whom presents with acute chest pain.

2. Case presentation

An 18-year-old teenager presented with complaint of left sided chest pain for five days. This was associated with significant loss of weight; 6 kg within two weeks and episodes of hemoptysis. Otherwise, he denied of having night sweat, fever, and dyspnea. No history of trauma to the chest. He is a college student and actively smokes e-cigarette.

His vital signs were stable and saturating well under room air. Lung examination revealed reduced breath sound over the left basal. Other system examinations were unremarkable. Chest radiograph showed multiloculated lesions over left lower lung zone indicating possibility of abscesses or cystic lesions. These lesions were causing mediastinal shift to the right (Fig. 1). Urgent computed tomography (CT) of thorax was performed and revealed large multi-cystic mass in the left inferior hemithorax causing mediastinal shift. Thin

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enhancing septations were seen with presence of air fluid level that indicative of abscess. This mass had no communication with tracheobronchial tree and the left lower lobe is almost completely collapsed by the mass (Fig. 2).

Based on the above findings, diagnosis of lung abscess was made with findings suggestive of intralobar bronchopulmonary sequestration as well. CT angiogram of thorax was performed to establish feeding blood supply and drainage of the pulmonary sequestration. The sequestrated lung received blood supply from thoracic artery and drained into left inferior pulmonary vein. Sputum specimen cultured pseudomonas aeruginosa. His final diagnosis was infected left intralobar bronchopulmonary sequestration with lung abscess. He received a total of 14 days duration of antibiotic and was referred to cardiothoracic surgeon for embolization.

3. Discussion

Bronchopulmonary sequestration, also known as pulmonary sequestration (PS) is a rare congenital pulmonary abnormality with incidence of 0.15%–6% [1,2]. It was first described by Rokitansky and Rektorzic over 100 years ago and the term sequestration was first used by Pryce. Multiple theories are proposed initially and it is now accepted that, PS is a separated lung tissues or lobe that has no communication with bronchopulmonary tree, with its own vascular supply [1,3]. There are two types identified; intralobar and extralobar sequestration. Intralobar is found to be more common and diagnosis usually made in late adolescent or early adulthood while extralobar are commonly diagnosed in fetal to infancy period.

Intralobar sequestration could be asymptomatic but when symptomatic, the most frequent reported symptom is recurrent chest infection, and only minority presents with chest pain [2,4,5]. According to a large retrospective study conducted in China, out of 2625 cases of PS, 1923 cases were symptomatic of which only 214 (11%) presented with chest pain [6]. Few recent small retrospective studies also showed similar findings of which, chest pain recorded to be the presenting symptom in about only 11–15% of their symptomatic intralobar cases [4,5,7]. Recurrent chest infection could be in a form of recurrent pneumonia, however, only few cases reported to have lung abscess [4,8,9]. Even though uncommon, clinical presentation of acute chest pain with suspected lung abscess as in this case should raise a suspicion of PS.

Radiological imaging is a mean to diagnose PS, besides histopathological confirmation. Certain chest radiograph abnormalities could suggest to this diagnosis. The simple intralobar PS may manifest as consolidation or homogenous opacity mostly at left post-erobasal area. This is persistent despite treatment. Other commonly reported chest radiograph changes are cystic or cavitary lesion with or without presence of air-fluid level within. This lesion could be singular or multiple and presence of air-fluid level does not indicative of infection but can also be lung abscesses [2,10]. The finding of cystic or cavitary lesions highly raised a suspicious of cystic malignancy as differential diagnosis while the consolidative changes raise a suspicion of lung mass. Other reported chest radiograph changes are mediastinal shift and pleural effusion [10].

The changes in chest radiograph help to illuminate the possibility of PS however, because of its diversity, CT angiogram of thorax is required to ascertain the diagnosis. The advantages are; a clearer evaluation of the lung parenchyma, ability to establish the non-association with tracheobronchial tree and the assessment of visceral pleura. Intralobar PS does not have its own visceral pleura while extralobar PS has a separate visceral pleura from the main lung. Furthermore, angiogram helps to assess the feeding arteries and

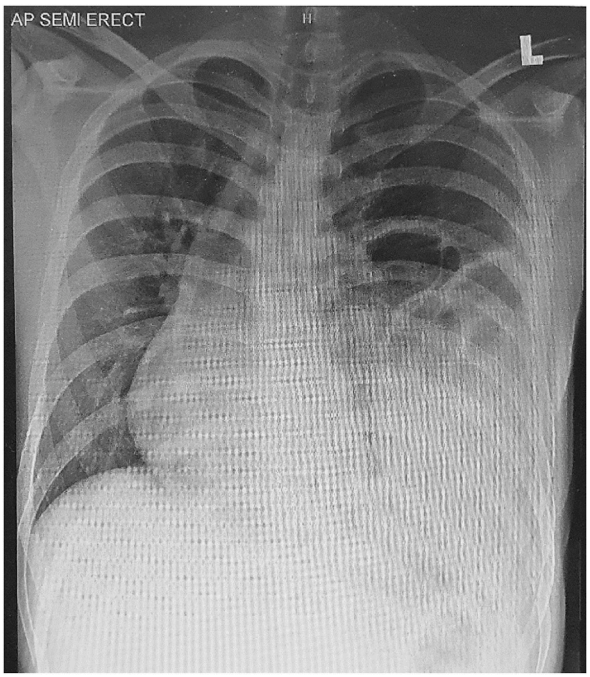

Fig. 1. Multi-loculated lesions seen over the left lower lung zone with mediastinal shift.

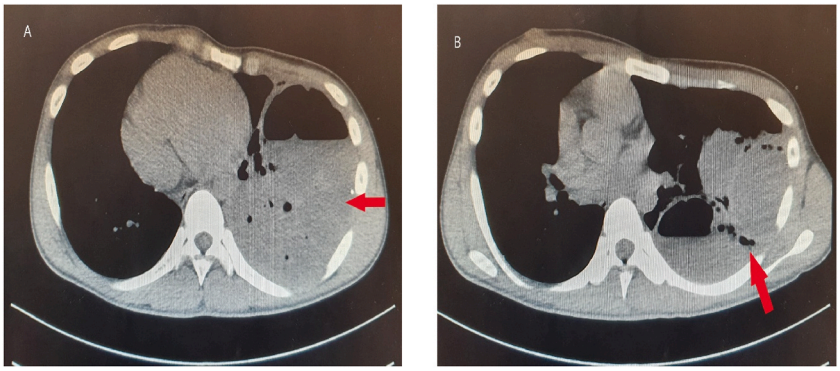

Fig. 2. Both A and B shows the multi-cystic mass at left inferior hemothorax with thin enhancing septations and presence of air fluid level within. (Red arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

draining veins of PS. This is essential for pre-operative assessment [2]. For intralobar PS, commonly reported feeding artery arises from thoracic aorta (73–76%) while it mostly drains into pulmonary vein [1,6,10,11]. For this case, patient's chest radiograph showed abnormalities that suggest PS; multi-loculated lung lesion at left basal with mediastinal shift and his CT angiogram of thorax helped with the diagnosis of intralobar PS by visualizing an abnormal lobe without connection with tracheobronchial tree and its own visceral pleura. This had branches of thoracic artery as the feeding artery and drained into pulmonary vein, as commonly reported. Treatment for pseudomonas lung abscess was based on CT finding of multi-cystic mass with thin enhancing septations and presence of air fluid level indicating abscess and sputum culture grew pseudomonas.

Current treatment options of intralobar PS are a surgical resection; lobectomy or sub-lobar resection, and embolization. Methods of surgery ranges from minimally invasive procedure such as video-assisted thoracoscopic surgery or conservative approach of thoracotomy [12]. There are few studies looking into the timing of operation and no definite conclusion has yet achieved. However, it is proposed that, the timing of operation should be performed soon after the diagnosis due to recurrent infection, risk of chronic lung parenchyma changes such as bronchiectasis, emphysema, and fibrosis, and risk of progression to malignancy [13]. Thus, in this case, early surgical treatment at the period of diagnosis was chosen in order to avoid the sequalae of PS as per evidence discussed.

4. Conclusion

Pulmonary sequestration is a rare pulmonary congenital malformation with intralobar PS being the more common sub-type. It commonly presents with recurrent pneumonia.

- An atypical chest pain in previously healthy young boy should raise a suspicion of PS possibility, together with support from radiological imaging.
- CT angiogram of thorax is a suitable modality to confirm PS and determine the vascular supply of the sequestrated lobe. This is helpful for approach in surgical treatment.
- PS leads to recurrent chest infection which in turn leads to chronic lung parenchyma changes and tends to progress to malignancy. Early surgical intervention after diagnosis is thought to help reduced the sequalae of PS.

Declaration of competing interest

There is no conflict of interest in the preparation of this manuscript.

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