Annals of Case Reports
Ghaly M, et al. Ann Case Rep: 7: 923.
www.doi.org/10.29011/2574-7754.100923
www.gavinpublishers.com

Case Report

Intralobar Bronchopulmonary Sequestration with Abscess in an Adult Female: A Case Report

Michael Ghaly, MD[1], Christian Motley[2], Kayla Nguyen[2*], Samuel Harris, MD[1]

[1]Baptist Memorial Hospital - North Mississippi Internal Medicine Residency, Oxford, MS, USA

[2]William Carey University College of Osteopathic Medicine, Hattiesburg, MS, USA

***Corresponding author**: Kayla Nguyen, William Carey University College of Osteopathic Medicine, Hattiesburg, MS, USA

Citation: Ghaly M, Motley C, Nguyen K, Harris S (2022) Intralobar Bronchopulmonary Sequestration with Abscess in an Adult Female: A Case Report. Ann Case Report 7: 923. DOI: 10.29011/2574-7754.100923

Received: 20 August 2022, **Accepted:** 24 August 2022, **Published:** 26 August 2022

Abstract

We report a case of bronchopulmonary sequestration with secondary abscess formation that resulted in a lower lobectomy of the right lung. A 32-year-old female with a productive cough and recurrent pneumonia was found to have a large complex intrapulmonary abscess in the right lower lobe secondary to an infected pulmonary sequestration. She was placed on antibiotics, underwent a chest tube placement for drainage of the abscess, and subsequently, she had a right lower lobe lobectomy. In patients with recurrent pneumonia, chronic productive cough, or persistent dyspnea on exertion, the diagnosis of bronchopulmonary sequestration should be considered and explored.

Keywords: Pulmonary abscess; pulmonary sequestration

Introduction

Bronchopulmonary sequestrations are masses of lung tissue that do not provide pulmonary function and have an arterial blood supply from the systemic circulation [1]. These lung tissue abnormalities are congenital. There are several theories on the pathogenesis of the formation of sequestrations. The most accepted theory involves the development of a lung bud during embryogenesis that is separate from the normal lung buds [2]. The separate lung bud then receives its own blood supply from the primitive splanchnic vessels [1]. There are two subtypes of sequestrations that are studied. They include intralobar and extralobar sequestrations. Intralobar sequestrations are contained within the same lung pleura of the adjacent lung tissue; whereas, extralobar sequestrations have their own pleura separate from adjoining lung tissue [2]. Intralobar sequestrations are by far the most common subtype, as they account for 75 to 90% of all bronchopulmonary sequestrations [3]. This type has an equal distribution of incidence between males and females [2]. Extralobar sequestrations are an estimated 10 to 25% of bronchopulmonary sequestrations, and there is a 4:1 male to female ratio of this type of malformation [1]. The extralobar sequestrations are more commonly to be accompanied by other congenital anomalies, for example, diaphragmatic hernias, bronchogenic cysts, congenital heart disease, or gastric duplication [4]. Because of the association of extralobar sequestrations and other congenital issues, they are usually diagnosed in the neonatal period, whereas intralobar sequestrations are diagnosed later in life [3]. This may be because of the overlap of clinical symptoms of intralobar sequestration with other pulmonary conditions like pneumonia, COPD, or bronchitis. The most common clinical presentation of patients with bronchopulmonary sequestration is recurrent pneumonia in a particular lobe of the lung [1]. Additional presentations may include persistent cough, shortness of breath, back pain, and hemoptysis [1]. The imaging of choice for diagnosing the sequestration along with the blood supply is CT Angiography [3]. In a study published in the European Journal of Cardio-thoracic Surgery, 71% of their patients with sequestration had blood supply from a single artery [2]. Blood supply for intralobar sequestrations arise commonly from the thoracic aorta, but there are documented cases of other origins [3]. The patient discussed was found to have an anomalous artery found at the level of the diaphragm that originated from the descending aorta.

Case

Our case is about a 32-year-old female with a past medical history of pulmonary embolism, reported pulmonary abscess vs pleural effusion requiring chest tube, recent COVID-19 pneumonia, and previous tobacco abuse who was transferred to us from another facility with initial complaints of shortness of breath and cough. Prior to admission, she had an increasing cough for five days that was productive of foul-smelling, thick, yellow to green sputum. She also reported nausea and left-sided chest pain that worsened with coughing and deep breathing. She denied fever, chills, abdominal pain, constipation, or diarrhea. The patient had similar symptoms one to two years ago requiring drainage of fluid via a chest tube at that time. Outside records were not able to be obtained, and it was unclear if this was an abscess or pleural effusion. On examination, the patient was alert and in no acute distress. Her lung examination revealed decreased breath sounds, wheezing, rhonchi, and rales to the right lung base. She was in no respiratory distress; however, she coughed persistently during the exam. Vitals on presentation to the outside facility showed that the patient was hypertensive at 145/70 mmHg, tachycardic with a heart rate in the 110s bpm, and febrile at 102.1 F (38.9 C). She was saturating at 100% on room air. The patient underwent a CT Angiogram (Figure 1) with and without contrast of the chest. The preliminary report showed a large, multiloculated fluid collection involving the right lower lobe (Figure 1). Connecting the patient's vital signs and pneumonia, she was diagnosed with sepsis due to a respiratory source, treated appropriately per sepsis guidelines, and transferred to our facility for higher level of care.

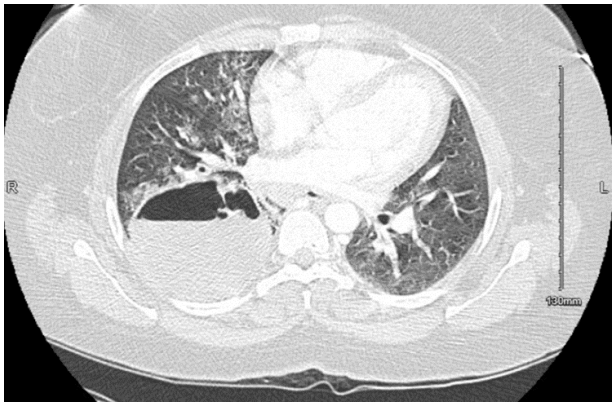

Figure 1: Occupying a majority of the right lower lobe is a large air-fluid collection measuring at least 11 cm craniocaudally by approximately 9 cm transverse by approximately 7.3 cm AP. Multiple fluid levels are seen throughout this area and the appearance is most consistent with large intrapulmonary abscesses. Associated areas of pneumonia are seen within the right lower lobe. Additionally, there is a relatively large feeding arterial vessel measuring 5-6 mm in diameter coursing into the right lower lobe, supplying this right lower lobe region including the large air-fluid collection.

Upon presentation to our facility, the patient was placed empirically on Unasyn and vancomycin. The initial CTA was reviewed again by a radiology resident from the previous institution. We were made aware of the final CTA report, which showed an area in the right lower lobe containing multiple fluid levels with associated areas of pneumonia and an arterial vessel coursing to and supplying this area, characteristic of a bronchopulmonary sequestration. The patient was also found to have bulky subcarinal and right hilar adenopathy. A chest tube was placed by Interventional Radiology for drainage of the abscess and Cardiothoracic Surgery was subsequently consulted. Per their recommendations for treatment of the sequestration, the patient was scheduled for a bronchoscopy in preparation for a lobectomy. The patient underwent a bronchoscopy six days after admission to rule out an endobronchial lesion. Bronchoscopy findings revealed a normal pharynx, and the vocal cords were normal in appearance and moved symmetrically with phonation. There were significant mucous plugs present, which were lavaged and suctioned until completely clear. The bronchial mucosa demonstrated changes consistent with mild chronic bronchitis. There were no observed endobronchial tumors. The following day, the patient underwent general anesthesia and a lower lobe lobectomy. A posterolateral thoracotomy was performed preserving the serratus anterior muscle. Upon entering the chest there were dense adhesions, which were lysed. The lung was mobilized superiorly and inferiorly to the level of the diaphragm. Upon reaching the medial portion of the diaphragm inferiorly there was a very large arterial branch which supplied the lower lobe. This was isolated, doubly tied with silk stitch and suture ligated with 4-0 Prolene. A segmentectomy was not possible because of the extent of the disease. In light of this a lower lobe lobectomy was performed as follows: The branches from the inferior pulmonary vein to the lower lobe were identified and transected. There was an anomalous branch from the inferior pulmonary vein which supplied the middle lobe, and this was preserved. The upper and middle lobes aerated well. Progel was used to seal small air leaks, with straight chest tubes placed (Figure 2). The patient was then taken back to the recovery room in stable condition. The rest of the patient's stay was uneventful. Blood and sputum cultures were negative for any growth. AFB with stain, and fungal cultures were also negative. However, the BAL culture grew *Streptococcus viridans*, and the nasopharyngeal culture grew methicillin-sensitive *Staphylococcus aureus*. She was discharged with 14 days of amoxicillin-clavulanate 875-125 mg, naproxen 500 mg, and oxycodone-acetaminophen 5-325 mg. The patient was unfortunately lost to follow up at our facility.

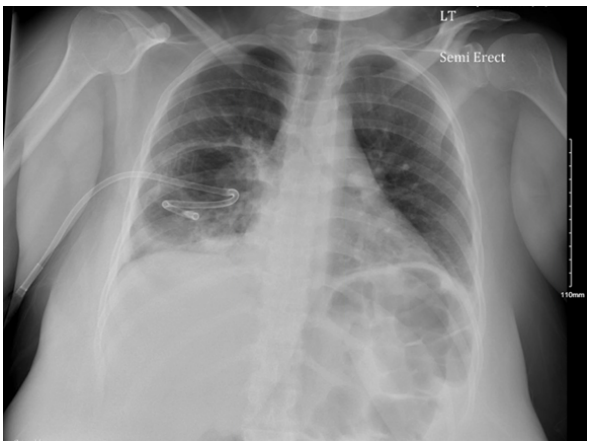

Figure 2: Right-sided chest tube in place with no pneumothorax. Very little fluid remains into the right lung abscess.

Discussion

Pulmonary sequestration is a rare congenital malformation that is characterized by lung tissue that receives blood supply separately from the source that is supplying the surrounding lung tissue [1]. Sequestrations are anatomically disconnected from the tracheobronchial tree [2]. This anatomical anomaly is thought to represent six percent of all lung malformations present at birth [1]. Clinical presentation of patients with bronchopulmonary sequestration aligns with many other lung conditions and can be difficult to diagnose. Patients may be asymptomatic. Others, however, could present with symptoms as we observed in this patient including recurrent pneumonia, hemoptysis, chest pain, shortness of breath, productive cough and wheezing [2]. The abnormal chest X-ray led to a CT angiography in order to confirm the pathology. As surgery is the treatment of choice, our patient had necessary surgical intervention which included a lobectomy with improvement in her symptoms. The age of diagnosis varies between the different types of sequestration, but most cases present under the age of twenty [1]. We present a case of a 32-year-old female with an undiagnosed bronchopulmonary sequestration following recurrent episodes of pneumonia and what was potentially diagnosed as a pulmonary abscess. Between the two types of sequestration, the patient in our case likely had an intralobar sequestration due to the lack of evidence on CTA or intraoperative findings showing separate pleura surrounding the sequestration.

Conclusion

In this case, we have a 32-year-old female with a previously undiagnosed intralobar bronchopulmonary sequestration suffering from periodic episodes of dyspnea and cough consistent with purulent sputum. Timely diagnosis coupled with targeted antibiotics and surgery can be a successful strategy in management and care. Following "incidental discovery on computed tomographic chest scans", recurrent pneumonia, as seen in our case, is the most common clinical presentation for an intralobar sequestration [1]. Swift identification, diagnosis, and proper treatment of lung tissue abnormalities can greatly improve quality of life for this patient population. The purpose of this paper is to emphasize that for any patient with recurrent pneumonia, chronic productive cough, or persistent dyspnea on exertion, a diagnosis of bronchopulmonary sequestration should be considered and explored.

References

1. Chakraborty RK, Modi P, Sharma S (2021) Pulmonary Sequestration. [Updated 2021 Jul 26]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing.
2. Cooke C (2006) Bronchopulmonary Sequestration. Respiratory Care June 51: 661-664.
3. Van Raemdonck D, De Boeck K, Devlieger H, Demedts M, Moerman P, et al (2001) pulmonary sequestration: a comparison between pediatric and adult patients. Eur J Cardiothorac Surg 19: 388.
4. Singh A, Mandelia A, Verma P, Naranje KM, Gupta G (2022) Giant feeding artery from abdominal aorta in intralobar pulmonary sequestration. J Indian Assoc Pediatr Surg 27: 270-272.