CASE SERIES

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Video-assisted thoracoscopic resection of intralobar pulmonary sequestration in adults

Kristi Pence, Puja Gaur, Edward Chan, Min P. Kim

ABSTRACT

Introduction: Intralobar pulmonary sequestration is a rare congenital abnormality that is usually diagnosed in childhood. When diagnosed in adults, patients are usually symptomatic and undergo open thoracotomy and lobectomy. However, video-assisted thoracoscopic surgery (VATS) has become a viable alternative. Case Series: We present three cases in which adults underwent VATS lobectomy. The resection of the aberrant vessels was aided by the superior visualization of VATS without any additional risk for the patients. All of the procedures were successfully completed without any major morbidity. Conclusion: Videoassisted thoracoscopic surgery (VATS) allows for superior visualization, decreased length of stay, decreased pain medication, and less morbidity than thoracotomy, and should be considered for management of adult patients with pulmonary sequestration.

Keywords: Pulmonary sequestration, Adult, Videoassisted thoracoscopic surgery (VATS), Lobectomy

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INTRODUCTION

Pulmonary sequestration is a rare congenital abnormality that consists of a non-functioning segment of lung tissue in which there is no communication between the sequestrated lung and the bronchial tree or pulmonary arteries. It is estimated to comprise 0.15-6.4% of congenital pulmonary malformations and approximately 1% of pulmonary resections [1]. In intralobar sequestration, the lung tissue lies within the same visceral pleura of the lobe and typically has pulmonary venous outflow, while extralobar sequestration has its own visceral pleura and has systemic venous outflow. The standard treatment for symptomatic intralobar sequestrations is a lobectomy, which is typically performed through an open thoracotomy. Recently, VATS lobectomy has grown in popularity. We present a series of three adults diagnosed with intralobar pulmonary sequestrations safely treated with VATS lobectomy.

CASE SERIES

Case 1

A 29-year-old male presented to the clinic with a 10year history of hemoptysis that had worsened over the



previous three months. Computed tomography (CT) showed a left lower lobe pulmonary sequestration with a 1.5 cm anomalous vessel arising from the thoracic aorta (Figure 1A). Left VATS was performed using a 4 cm utility incision placed in the 4th intercostal space in the mid-axillary line, a 1-cm camera port placed in the 7th intercostal space posterior to posterior axillary line and a 1-cm utility incision was placed in the 7th intercostal space in the mid-axillary line. The aberrant vessel coming off the aorta was isolated (Figure 1B) using electrothermal bipolar tissue sealing system and divided by vascular stapler. Three small branches from pulmonary artery to left lower lobe were identified and divided. The patient's pain was well controlled and he was discharged on postoperative day-3 with resolution of his hemoptysis. At follow-up one month later, he had resolution of hemoptysis.

Case 2

A 17-year-old boy with an autism spectrum disorder and a history of recurrent left lower lobe pneumonia for five years underwent a CT scan that showed a complex cavitary lesion (9.3x7.1 cm) in the left lower lobe with an air-fluid collection and a 4-mm vessel arising from the descending thoracic aorta extending to the left lower lobe consistent with a left lower lobe intralobar pulmonary sequestration. After a course of Zosyn for four days, the abscess decreased in size. Patient then underwent left VATS lower lobe lobectomy, as discussed in detail in Case 1. Two small anomalous vessels were noted to arise from the aorta going to left lower lobe, in addition to a small branch from pulmonary artery to superior segment of left lower lobe. The patient's pain was controlled without narcotics. Repeated chest X-rays showed increasing left apical pneumothorax despite chest tube remaining in place, so he had an IR-placed chest tube placement. On post-operative day-5, chest X-ray showed no pneumothorax so both chest tubes were discontinued. He was discharged home on postoperative day-6 with an additional four weeks of augmentin and Doxycycline. He was discharged on postoperative day-6. Upon his follow up visit one month later, the patient's pneumonia had resolved.

Case 3

A 37-year-old female presented to her primary care provider with complaints of dyspnea and coughing that had started during a pregnancy the previous year. She had suffered from severe GERD during pregnancy, which resolved after her child was born, but she continued to have a persistent cough and intermittent low fevers. A CT scan of her chest without IV contrast showed that her right lower lobe had bronchiectasis. A bronchoscopy showed irritation of mucosa and a small pocket of pus in the right lower lobe and the washings were negative for malignancy. After course of levaquin for seven days, repeat CT scan

with IV contrast showed right lower lobe intralobar pulmonary sequestration with associated abscess (Figure 1C) supplied by 6 mm branch from descending aorta just superior to the diaphragm. The patient underwent right VATS lower lobectomy. Due to the low location of the anomalous branch from the descending aorta, the posterior axillary line incision placed at the 8th, instead of 7th, intercostal space. The 6 mm anomalous branch from the descending aorta just superior to the diaphragm was isolated and divided (Figure 1D). The culture grew *Staphylococcus aureus* and patient was treated with Levaquin for five days. Her pain was well controlled and she was discharged on postoperative day-2. At follow-up one month after resection, she had resolution of her cough and low-grade fevers.

DISCUSSION

Pulmonary sequestration is a rare congenital anomaly that usually manifests early in life, but can be diagnosed as an adult. Symptoms are present in 84% of adults diagnosed with pulmonary sequestration and 71–79% of sequestrations are in the left lower lobe [2, 3]. A retrospective analysis of 2625 patients showed that the most common symptoms of pulmonary sequestration in adults are cough (69%), fever (39%), hemoptysis (28%), and chest pain (11%), with 13% of patients being asymptomatic [3]. Chest X-rays often simply show

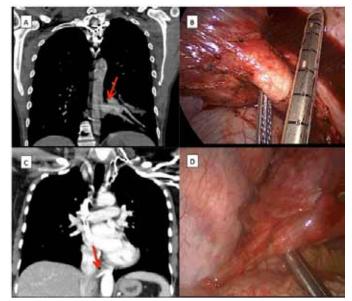


Figure 1: Aberrant vessel to pulmonary sequestration (A) A computed tomography image shows a 1.5 cm vessel arising from the descending aorta going into the left lower lobe intralobar pulmonary sequestration, (B) A thoracoscopic image of an aberrant blood vessel arising from the aorta with vascular stapler going around the isolated vessel, (C) A computed tomography image shows a 6 mm vessel arising from descending aorta, and (D) A thoracoscopic image of an aberrant blood vessel in the inferior pulmonary ligament isolated by thoracoscopic instrument.



bronchiectasis or a hazy opacity. Chest CT scans show mass lesions (49%), cystic lesions (29%), cavitary lesions (12%), and pneumonic lesions (8%) [3, 4]. The lack of specific symptoms and radiographic findings can mimic a variety of clinical conditions.

The majority of lobectomies are still performed via thoracotomy, but the prevalence of VATS resection has increased in recent years. Interestingly, VATS resection is utilized more often in children than adults; 75% of lobectomies for sequestration in children are performed via VATS compared 16% in adults [5]. In our series, two patients had a long-standing history of recurrent symptoms, resulting in significant scar tissue, abscess cavities and less than ideal tissue planes. In our experience, VATS allows for better visualization of the planes and aberrant vessels, making them less susceptible to injury to the vasculature or lung parenchyma.

A thorough review of the blood supply to the symptomatic portion of lung can aid in diagnosis, as anomalous arteries arise from the thoracic aorta (81–86%), abdominal aorta (7–19%), and phrenic artery (5.6%) [3, 4]. The majority of sequestrations are supplied by a single artery, but 21% are supplied by two or more arteries [3]. The size, number, and origin of the supplying artery are highly varied. The correct identification of these branches is of vital importance to avoid vascular injury. VATS is able to provide excellent visualization of the vasculature, thus providing a safe way to identify and control the vessels.

CONCLUSION

All of our patients successfully underwent videoassisted thoracoscopic surgery (VATS) lobectomy, despite dense adhesions and abscess cavities, multiple aberrant vessels, and difficult to access anatomy, with no postoperative complications. Additionally, our patients' pain was adequately controlled with oral pain medications. VATS lobectomy is a safe and viable option for pulmonary sequestration resection and should be considered for treatment of pulmonary sequestration.

Author Contributions

Kristi Pence – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Puja Gaur – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published Edward Y. Chan – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Revising it critically for important

intellectual content, Final approval of the version to be published

Min P. Kim – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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