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Case Series

Endovascular embolization for pulmonary sequestration in adults: An adjunctive technique to delay surgical intervention

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ABSTRACT

Pulmonary sequestration is a congenital malformation characterized by a non-functioning segment of dysplastic lung that lacks communication with the bronchial tree and contains an aberrant systemic arterial supply. Pulmonary sequestration is classically managed surgically; however, surgical intervention is not always feasible on an emergent basis. We present two cases, in support of recent literature, that reinforce the use of transcatheter embolization to achieve hemostasis and delay surgical intervention.

Keywords: Pulmonary sequestration, Embolization, Endovascular, Case series, Adult

INTRODUCTION

Pulmonary sequestration is a congenital malformation characterized by a non-functioning segment of dysplastic lung that lacks communication with the bronchial tree and contains an aberrant systemic arterial supply. Pulmonary sequestrations have been reported to represent a mere 1–6% of all congenital pulmonary anomalies.[1] Nearly two-thirds of sequestrations are intralobar, sharing a common pleura with normal lung, and these cases often presenting in adolescence or in older children.[2] The less common extralobar variant with a separate pleural lining presents earlier, typically in neonates.[2] Depending on the type of sequestration and the volume of lung mass that the malformation comprises, clinical presentation ranges from asymptomatic to symptomatic presentations with hemoptysis, pulmonary infection, or persistent cough.[3]

Conventionally, both extralobar and intralobar sequestrations are treated surgically. Extralobar sequestrations can usually be resected while sparing normal lung tissue due to their separate pleural investment. Intralobar sequestrations typically require segmental resection or lobectomy through video-assisted thoracoscopic surgery, robotic-assisted thoracoscopic surgery, or open thoracotomy and thus have a higher risk for complications. Complications of surgical intervention including chylous leak, intraoperative bleeding, chronic chest pain, arm numbness, and pneumonia have been reported in up to 28% of cases, with the most significant complications being empyema, hemothorax, and hemopneumoperitoneum.[2,4] There are also clinical scenarios where emergent surgical intervention presents significant risk of mortality, such as in patients at risk of life-threatening hemorrhage with a source of bleeding that is difficult to

control.[5] Emergent endovascular embolization has emerged as an option to achieve hemostasis and allow delayed surgical intervention. Notably, case reports of successful embolization have now been described in adults.[1-11] We report two cases of endovascular embolization that further illustrates the utility of emergently stopping hemoptysis, allowing for delayed surgical intervention for pulmonary sequestration.

CASE REPORT

Two recent cases of intralobar pulmonary sequestration were reviewed along with demographics, medical history, and outcomes [Table 1]. Post-embolization angiogram demonstrated stasis in both cases. Post-embolization clinical status, operative findings, and post-operative course were reviewed.

Case 1

The patient was a 36-year-old female who presented to the emergency department with cough and hemoptysis (approximately 100 cc) for 1 day. The patient noted three prior episodes of hemoptysis that occurred over the previous 2 months that resolved within 24 h on each occasion without seeking medical attention. In the emergency department, she had an initial blood pressure of 129/78 mmHg, heart rate of 102 bpm, and oxygen saturation of 96% on room air. Initial hemoglobin concentration was 10.4 g/dL. Pre-procedural computed tomography angiography revealed right lower lobe pulmonary sequestration with aberrant systemic arterial supply arising off the celiac trunk [Figure 1]. Embolization was performed on the day of admission through a 5 French Imager II Bern catheter (Boston Scientific, Marlborough, Massachusetts) using 11 Interlock-35 Fibered IDC occlusion system coils (one 4 mm × 10 cm, two 6 mm × 10 cm, two 6 mm × 20 cm, two 8 mm × 40 cm, two 10 mm × 40 cm, and two 12 mm × 40 cm) (Boston Scientific, Marlborough, Massachusetts).

The patient had immediate resolution of hemoptysis following embolization. The patient subsequently developed pain, low-grade fever, and tachycardia the night following the procedure; however, the patient's white blood cell count remained within normal limits. A CTA of the patient's chest was performed 2 days post-embolization, which showed progressive atelectasis of the right lower lobe. Robotic-assisted right lower lobectomy was performed 5 days post-embolization. Intraoperative blood loss was 50 cc. Pathology showed partial necrosis associated with the sequestered segment. The patient was discharged uneventfully 3 days post-lobectomy.

Case 2

The patient was a 34-year-old male with no prior medical history who presented to the emergency department with 1 day of hemoptysis (approximately 150–200 cc). Initial heart rate was 105 bpm, blood pressure was 204/136 mmHg, and oxygen saturation was 96% on room air. During the first 24 h of admission, the patient continued to have intermittent episodes of hemoptysis totaling approximately 200 cc in volume; however, his blood pressure stabilized with medical management and his oxygenation remained stable on room air. Pre-procedural computed tomography angiography revealed left lower lobe pulmonary sequestration with aberrant systemic arterial supply arising directly off the descending thoracic aorta [Figure 2]. Embolization was performed approximately 24 h after admission through a 5 French Mickelson catheter (AngioDynamics Inc., Latham, New York) and Renegade High-Flow microcatheter (Boston Scientific, Marlborough, Massachusetts) using Surgifoam Absorbable Gelatin Sponge admixed with contrast as a slurry (Ethicon Inc., Somerville, New Jersey).

The patient had immediate resolution of hemoptysis following embolization. The patient was taken for resection of the sequestration the day after embolization. Robotic-assisted lobectomy was attempted; however, the patient had dense adhesions requiring tedious lysis that necessitated conversion to open thoracotomy. Intraoperative blood loss was 400 cc. The patient's post-operative course was unremarkable, and he was discharged 10 days following resection.

DISCUSSION

Our cases illustrate the utility of emergent transcatheter embolization for pulmonary sequestration to manage active hemorrhage and delay surgical intervention. Surgical

Table 1: Demographics, medical history, presenting symptoms, and surgical findings.

Case	Age	Gender	Location of sequestration	Medical history	Presenting symptoms	Post-embolization	Surgical findings
1	36	Female	Right lower lobe	None	Hemoptysis	Low-grade fever (38.2 C), tachycardia (111 bpm), right thoracic pain	Ischemic necrotic portions of RLL post-lobectomy
2	34	Male	Left lower lobe	None	Hemoptysis	Asymptomatic	No reported necrotic portions

RLL: Right lower lobectomy

resection has long been the standard of care for pulmonary sequestrations. However, emergent surgical intervention presents significant complications and is highly dangerous in patients with a difficult to control aberrant blood supply.[1-2,5] Controlling bleeding intraoperatively can be a challenge because the systemic arterial supply of the sequestered lung tissue has a higher blood pressure than a normal pulmonary artery, and any unintentional damage to the aberrant artery can result in life-threatening hemorrhage.[6]

Recently, pre-operative management through transcatheter embolization has emerged as an effective option in adults. The current literature suggests pre-operative embolization of the major arterial supply to the sequestered lung segment may lead to reduced intraoperative blood loss, with smaller sequestrations responding better to embolization.[7,8]

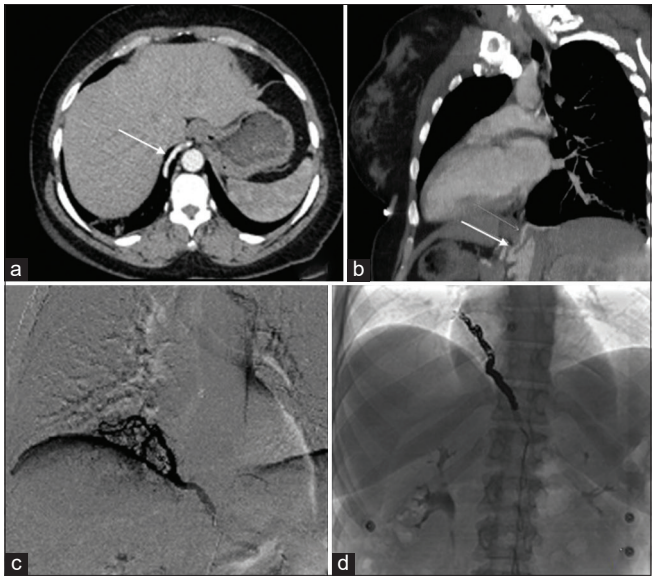

Figure 1: A 36-year-old female who presented with large volume hemoptysis. Axial arterial phase (a) and curved multiplanar (b) CT demonstrating an aberrant single vessel extending superiorly from below the diaphragm (arrows). (c) Digital subtraction angiography demonstrating an aberrant artery off the celiac trunk supplying the right lower lobe sequestration. (d) Post-coil embolization image demonstrating appropriate coil pack.

Intraoperative blood loss during both robotic- and video-assisted lobectomy is lower than open thoracotomy. The previous studies have reported mean blood loss of 87 cc and 170 cc during robotic- and video-assisted lobectomy, respectively; however, in patients with dense pleural adhesions, intraoperative blood loss is higher and can range from 200 to 500 cc.[9,10] In comparison, a reduction in intraoperative blood loss, 50 cc, was observed during robotic-assisted lobectomy post-embolization in Case 1. The 400 cc of intraoperative blood loss observed in Case 2 is more difficult to evaluate due to the patient's dense pleural adhesions and conversion to open thoracotomy, but it is likely that pre-operative embolization still provided intraoperative benefit due to the complicated nature of the case.

Transcatheter embolization has been described as a stand-alone intervention for pulmonary sequestration that may obviate the need for surgical intervention in some patients; however, embolization alone carries risk of incomplete occlusion of arterial supply and subsequent collateral formation, which could lead to recurrent infection or persistent perfusion to the sequestration.[7] Additional concerns include inadvertent embolization of non-targeted arteries secondary to migration of embolization material.[10] Surgical resection following embolization likely reduces long-term complications, risk of infection, or other pathology.[8,12,13]

Multiple different embolization methods were considered for each case. There is no established consensus for embolization method in pulmonary sequestration; the use of coils, calibrated particles/microspheres, Gelfoam, absolute alcohol, vascular plugs, and n-butyl cyanoacrylate have all been described in the recent literature.[11,14] The intent for both patients was surgical resection following embolization, and thus, embolization method was ultimately chosen according to surgeon preference. When choosing the pre-operative embolic agent for pulmonary sequestration, it is important to consider both the aberrant artery caliber and location of origin.[15] Multiple case reports support the use of coils in small, tortuous, or branched vessels.[16] However, operative dissection of an aberrant artery originating from

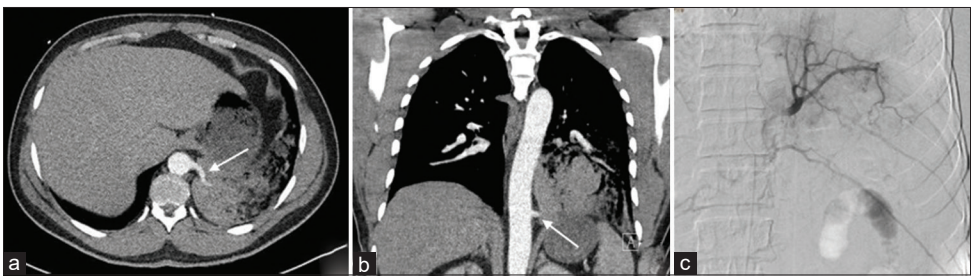

Figure 2: A 34-year-old male who presented with large volume hemoptysis. Axial (a) and coronal (b) arterial phase CT demonstrating a large single vessel off the descending thoracic aorta supplying the left lower lobe (arrows). (c) Digital subtraction angiogram demonstrating the aberrant arterial feeding vessel off the thoracic aorta.

the thoracic aorta can be technically challenging if pre-operative embolization is performed with an intravascular coil, as location of the intravascular coil must be identified a during surgical dissection and arterial cross-clamping.[15] This factor influenced the decision to embolize with Gelfoam in Case 2. In Case 1, the decision to embolize with coils was consistent with the few case reports that describe successful pre-operative embolization of aberrant arterial supply originating from below the diaphragm.[17,18] Ensuring adequate pre-operative hemostasis is especially important when the aberrant supply originates below the diaphragm, as the bleeding point can be lost deep to the diaphragmatic hiatus intraoperatively.[17]

CONCLUSION

Embolization is an effective temporizing therapy for patients presenting with active hemoptysis secondary to pulmonary sequestration. Embolization also serves as a safe bridging therapy to surgical intervention and may decrease intraoperative blood loss at the time of pulmonary sequestration resection. Operators performing pre-operative embolization for pulmonary sequestration should carefully consider embolic agent based on aberrant artery characteristics and location of origin.

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Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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