Robotic right lower lobectomy for a persistent large pulmonary arteriovenous malformation following repeated coil embolization

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ABSTRACT

Pulmonary arteriovenous malformations create continuous shunting of unoxygenated blood through the lungs into the systemic circulation. These malformations are asymptomatic if small, but cause serious symptoms as they grow in size. Treatment primarily consists of endovascular embolization; lobectomy is preserved for recurring or endovascularly untreatable cases. We describe a case of a 24-year-old man who was first treated with coil embolization 10 years previously, with complete symptom resolution. However, more recently he noted recurrent exercise intolerance, with shortness of breath and hypoxemia. After repeat re-embolization, a computed tomography scan noted some persistent flow. Given the patient's young age, we considered resection as a definite therapy. The patient underwent an uncomplicated robot-assisted right lower lobectomy. Afterward, his symptoms resolved completely. In selected cases, robotic lobectomy for pulmonary arteriovenous malformation is feasible and safe. (J Vasc Surg Cases Innov Tech 2024;10:101605.)

Keywords: Arteriovenous malformation; Embolization; Coils; Robotic; Pulmonary lobectomy

Pulmonary arteriovenous malformations (PAVMs) are abnormal direct connections between the pulmonary artery (PA) and vein that result in a right-to-left shunt. They can manifest as single focal or multiplex lesions. PAVMs are strongly associated with hereditary hemorrhagic telangiectasia (HHT); approximately 70% of PAVM cases are associated with HHT, and 15% to 35% of patients with HHT have PAVMs. The male-to-female ratio is approximately 1:1.5 to 1:1.8.[1] In a Japanese population computed tomography (CT) study, the prevalence was estimated at 38 per 100,000 individuals.[2] Although most AVMs are asymptomatic, the occurrence of symptoms correlates with the size of the lesion. Typical symptoms include dyspnea, exercise intolerance, migraine, brain abscess formation, rarely bleeding and even transient ischemic attack or stroke through paradox embolization. Hypoxemia and consequently polycythemia can be present.[3-5] CT angiography (CTA) is considered the gold standard in the diagnosis, and contrast-enhanced echocardiography is best for screening.[6,7]

Currently, there is no vascular surgical guideline for the preferred treatment of PAVMs, although according to other guidelines, embolization is considered the first choice over surgical interventions.[7,8] Indications for treatment include symptoms of hypoxemia, the presence of a feeding artery ≥2 mm, and the prevention of neurological complications and lung hemorrhage.[1,3,6,7] Coils or vascular plugs are considered safe and effective in treating PAVMs; however, long-term follow-up is advised, because recanalization or enlargement of untreated feeding branches can occur.[3,9,10]

Pulmonary resection is preserved generally in cases that are untreatable with an endovascular approach. To the best of our knowledge, there has been only one previous report of robot-assisted lobectomy for recurrent PAVM.[11] The patient provided informed consent to the report of his case details and imaging studies.

CASE REPORT

We report the case of a 24-year-old man with a history of PAVM in the right lower lobe, which was previously coil embolized when he was 14 years old. HHT was ruled out with genetic testing. He underwent routine yearly follow-ups with chest CTA every 2 to 3 years. In 2023, he presented with slowly progressive shortness of breath on exertion. Physical examination revealed no abnormalities, with a slightly decreased resting oxygen saturation of 96% and a slightly increased red blood cell count of 5.99 million per microliter, suggesting mild chronic hypoxemia. CTA confirmed a 4.4 × 3.7 × 3.5-cm arteriovenous malformation in the right lower lobe (Fig 1).

After obtaining informed consent, we attempted to embolize the malformation inflow (Supplementary Video 1, online only). The right femoral vein was accessed, a 4F sheath was

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Additional material for this article may be found online at www.jvscit.org.

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The editors and reviewers of this article have no relevant financial relationships to disclose per the Journal policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

2468-4287

https://doi.org/10.1016/j.jvscit.2024.101605

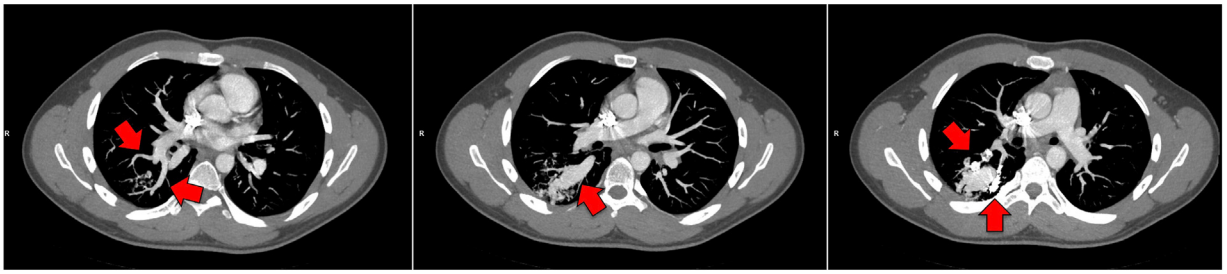

Fig 1. Computed tomography angiography (CTA) images before the second embolization attempt. **(Left)** Two major feeding branches. **(Middle)** Enlarged pulmonary vein, which forms as the outflow. **(Right)** Previously inserted coils.

introduced, and the PA was cannulated. After performing diagnostic angiography to confirm the location of the malformation, the sheath was upsized to a 90-cm-long 6F device, that was parked at the right interlobar PA. The distal branches of the right PA were cannulated using an 0.018-inch wire and a microcatheter. After confirming the feeding branches with selective angiography, including a previously treated branch that showed recanalization, detachable coils were placed and completion angiography was performed, confirming diminished flow in the AVM.

We administered 14,000 U Na-heparin in fractions during the procedure (10,000, 2000, and 2000) and the activated clotting time (ACT) was monitored to a target between 250 and 300 seconds (ACTs of 317, 265, and 267 seconds were measured throughout.) At the end of the procedure, 50 mg of protamine was given.

The postoperative course was uneventful, and no complications were noted (Fig 2). Three months after the procedure, a follow-up CTA was performed, which showed continuous filling of the AVM. The patient remained symptomatic and complained of ongoing shortness of breath on exertion (Fig 3).

Considering the previously unsuccessful endovascular attempts and adequate pulmonary function (forced expiratory volume in 1 second [FEV_1] of 100%; diffusion capacity of the lungs for carbon monoxide [$DLCO_2$] 81% of predicted), the patient underwent robot-assisted right lower lobe resection (Supplementary Video 2, online only). After general anesthesia and double-lumen intubation, the patient was placed in the left lateral decubitus position. The patient was prepped and draped and standard four robotic ports were placed. The first port was placed posteriorly by the scapula tip, the middle port in the posterior axillary line, and the third port in the anterior axillary line. Additionally, a robotic port was placed in the ninth intercostal space in the line of the scapula tip.[12] The camera was placed in the port at the posterior axillary line. The first port posterior to the scapula was used for retraction, and the other two ports were used for dissection and stapling. We released the inferior pulmonary ligament, divided the inferior pulmonary vein using a vascular robotic stapler, dissected the posterior fissure, and divided it with a robotic blue load stapler. Subsequently, we isolated and divided the basilar and superior segmental PA using a vascular robotic stapler. The anterior fissure was divided using a robot blue load stapler, and the right lower lobe bronchus was divided using a robot green load stapler. A leak test showed no air leak at the staple line and the right lower lobe was removed in a bag through the enlarged anterior incision.

After an uneventful postoperative course, the patient was discharged on the first postoperative day without complications. At 1 month follow-up, the patient had resolution of dyspnea on exertion with a blood oxygen saturation level of 100% in room air. The pathology report confirmed the presence of AVM in the removed right lower lobe without any additional findings. Subsequent follow-ups will be conducted through the patient's primary care physician, with specialist visits as indicated.

DISCUSSION

Although embolization is considered the first-line treatment for PAVMs, no randomized studies have supported this finding. It has become the preferred method owing to its minimally invasive and repeatable nature. It gained popularity in the 1980s, when it replaced the traditional open surgical approach. Since then, minimally invasive procedures have evolved, including video-assisted thoracoscopy and robotic surgery.

Although embolization procedures have high immediate success rates, recurrence is reported to be 20% to 35% over long-term follow-up.[1,9-11] This means that patients require long-term attention, repeated diagnostic scans, and possibly repeated interventions. For a younger patient, the cumulative effect of radiation exposure owing to repeated follow-ups and interventions can pose long-term quality of life and health concerns.[13]

Surgical removal of the affected pulmonary lobe is a definitive solution without the need for repeated interventions. The question is whether it has comparable perioperative results and a low complication rate as the current standard endovascular approach. Possibly, not for every case, but for a select group of patients, it might be a better alternative. A comparison of robotic-assisted lobe resections with open lobectomy and video-assisted thoracoscopy showed that patients who were operated on with the robot had fewer postoperative

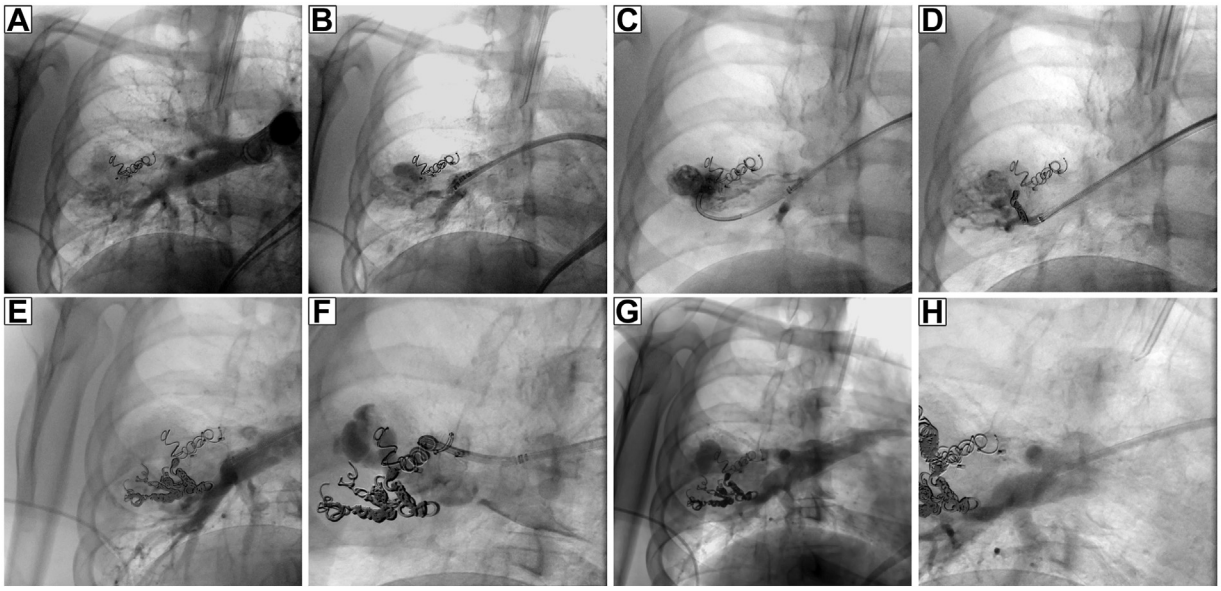

Fig 2. Steps of the coil embolization. **(A-C)** Previously placed coils can be seen in the right lower lobe superior segmental pulmonary artery (PA) branches, as well as filling of the pulmonary arteriovenous malformation (PAVM) from the right lower lobe basal segmental PA branches. Completion angiography shows diminished filling of the PAVM after selective coil embolization **(D** and **E)**. **(F** and **G)** Persistent filling of the PAVM can be appreciated through the previously (10 years before) embolized superior segmental branches. **(H)** Diminished filling after re-embolization of these previously treated vessels.

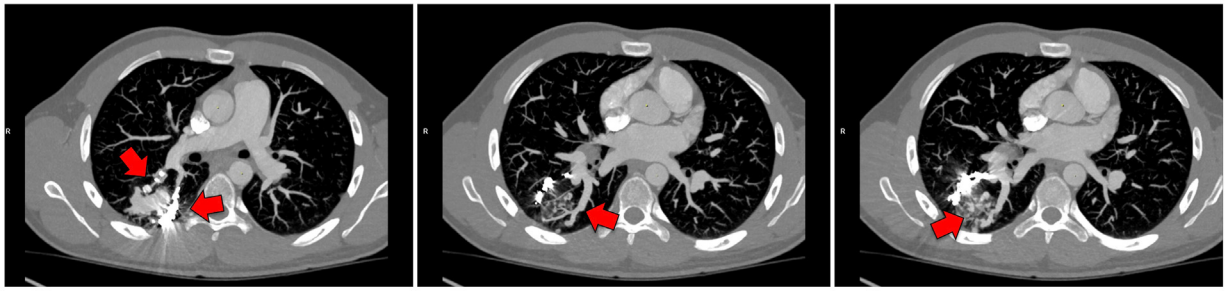

Fig 3. Follow-up computed tomography angiography (CTA) after the second embolization. The pulmonary arteriovenous malformation (PAVM) showed continuous filling despite multiple feeding branches that had been embolized.

complications and shorter lengths of stay than those undergoing other surgical approaches.[14] Because PAVMs are rare and mostly treated with embolization, there are no pooled data available regarding robotic-assisted lobectomy in this patient group. There has been no direct comparison between these two approaches either. There is only one previous case report on this topic.[11]

In our case, a young patient presented with a recurring PAVM, for which we elected to undergo re-embolization according to the recommendations in the literature.[7,8] The aim of embolization in this patient group is to close the feeding vessel as close to the nidus as possible to prevent collateral reperfusion and the occlusion of normal vessels preserving functional lung parenchyma. We chose to use detachable coils because most of the distal inflow vessels were small in diameter. Coil embolization is advantageous in this case because of the ease of use, the coil's adaptation to the vascular structure, and their smaller diameter, more flexible delivery system facilitating the reach, and occlusion of more distal feeding branches. Amplatzer vascular plugs require a larger diameter and a more rigid delivery system, which limits their use in distal embolization; they are used mostly to treat larger diameter, central feeding branches.[5,15] The use of microvascular plugs is another alternative; however, their use for PAVMs is less well-studied and microvascular plugs are generally suitable for more simple malformations, with fewer feeding vessels.[15,16]

Despite the promising early results of our attempt, the patient remained symptomatic, and a follow-up CTA confirmed the continuous filling of the AVM. Considering previous unsuccessful interventions, the size of the AVM, and the age of the patient, we chose to offer a more invasive, but possibly more durable, solution. The patient experienced no complications, and the length of stay (1 day) was comparable with that of endovascular solutions.

For selected patients with PAVMs, robot-assisted lobectomy can serve as an alternative to an endovascular approach. With the rapidly growing thoracic experience in robotic resection, this therapy should be considered increasingly as definitive. Therefore, when considering treatment options in patients where the endovascular approach seems challenging, the inclusion of a thoracic surgeon with robotic experience in the decision process should be considered.

CONCLUSIONS

In patients with recurrent PAVM or after multiple endovascular attempts, robotic-assisted lobectomy presents a viable minimally invasive option. More studies are required to determine whether this approach should be considered as a first-line definitive treatment option in selected patients.

DISCLOSURES

M.P.K. consults for Intuitive Surgical, Medtronic, Olympus, and AstraZeneca. A.B.L. receives research support from W. L. Gore & Associates; consults with Boston Scientific, W. L. Gore & Associates, and Siemens; and is a shareholder in Hatch Medical.

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Submitted Jul 1, 2024; accepted Aug 2, 2024.

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