Isolated endovascular repair of anomalous systemic arterial supply to the left basal lung

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ABSTRACT

Anomalous systemic arterial supply to the left basal lung is a rare congenital lung malformation, and its optimal treatment strategy is not well defined. We present a case of a 61-year-old man who underwent thoracic endovascular aortic repair (TEVAR) for anomalous systemic arterial supply to the left basal lung complicated with aneurysmal dilatation of the aberrant feeding artery. Computed tomography angiography after TEVAR revealed significant shrinkage of the aneurysmal portion as well as complete occlusion of the aberrant feeding artery. TEVAR proved to be a safe and efficient treatment for this rare arterial abnormality. (J Vasc Surg Cases Innov Tech 2021;7:481-3.)

Keywords: Anomalous systemic arterial supply to the left basal lung; Pulmonary sequestration; Aneurysmal dilatation; Feeding artery; Thoracic endovascular aortic repair

Pulmonary sequestration (PS) is uncommon congenital malformation of the lungs characterized by nonfunctional lung tissue separated from the normal tracheobronchial tree, and receives blood supplied from an aberrant systemic artery. PS is divided into two groups: intralobar PS (ILS) and extralobar PS, based on its relationship with the visceral pleura. Also, PS accounts for 6.4% of all congenital lung malformation, and the incident rate of ILS has been reported to be six times higher than that of extralobar PS.[1-4] We present a case with anomalous systemic arterial supply to the left basal lung (Pryce type I of all ILS), anatomically characterized by an aberrant feeding artery originating from the aorta, which supplies the basal segment as well as the missing sequestrated lung. Contrary to the conventional surgical treatments, including division of the anomalous feeding artery and segmental lobectomy, isolated thoracic endovascular aortic repair (TEVAR) was performed to simply occlude the aberrant feeding artery to treat this anatomical disorder. Informed consent for this case report was obtained from the patient.

CASE REPORT

A 61-year-old man was referred to our institute because of chest discomfort and an abnormal shadow of the hilar region on his chest X-ray. Enhanced computed tomography (CT) of the chest and aorta revealed an aberrant feeding artery to the left basal lung that originated from the descending thoracic aorta, and the arterial aneurysm was identified at the orifice of the aberrant artery, with a maximum diameter of 30 mm (Fig 1, *A* and *B*). There were no additional feeding arteries besides the aberrant artery from the systemic supply. The left pulmonary artery, its branches, and the bronchus were normal. The patient had no apparent history of recurrent pneumonia and congestive heart failure. Therefore, we decided to treat the patient by TEVAR without lung resection. Under general anesthesia in a supine position, the right common femoral artery was directly exposed. The presence of an aberrant aneurysmal feeding artery originating from the descending thoracic aorta was confirmed on the aortography (Fig 2). After systemic heparinization, TEVAR was performed to cover the orifice of the aberrant artery. As the aortic diameter of the landing zone was 28 to 29 mm in the proximal and distal site, a single piece of 34-34-100 mm GORE C-TAG endograft (W. L. Gore & Associates, Flagstaff, Ariz) was delivered and placed. The patient had an uneventful clinical course and was discharged on postoperative day 5. The enhanced CT, 1 month after TEVAR, demonstrated significant shrinkage of the maximum short diameter from 30 to 19 mm and the complete occlusion of the aberrant feeding artery (Fig 3).

DISCUSSION

Approximately 60% of cases with anomalous systemic arterial supply to the left basal lung demonstrate some clinical symptoms, such as bloody sputum, hemoptysis, fever, and chest discomfort.[1] Operative indications of overall PS still seem to be controversial.[3,4] However, in this disorder, massive bleeding due to ruptured pulmonary arteries, caused by persistent pulmonary hypertension and pulmonary arteriovenous fistula, is really a matter of concern if untreated. Therefore, surgical or interventional procedures are considered to be primary treatments of choice. Based on previous literature, treatment options are as follows: simultaneous division or

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Author conflict of interest: None.

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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.2021.06.005

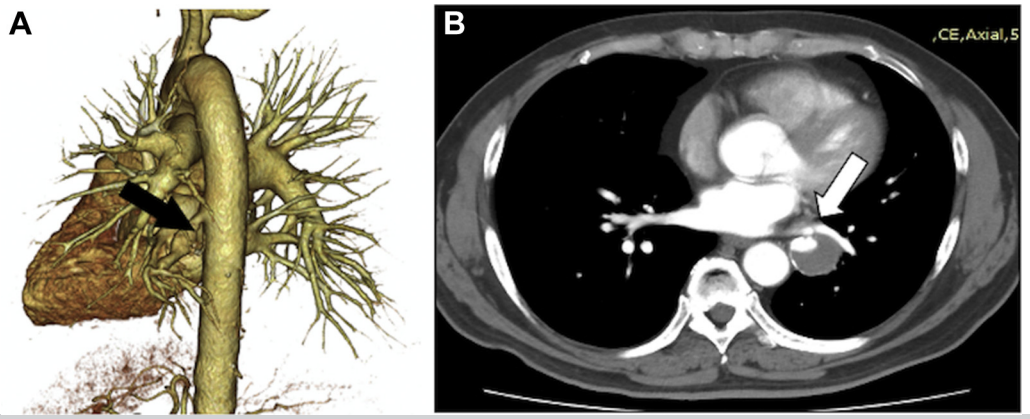

Fig 1. Preoperative three-dimensional computed tomography (CT) reveals that an aberrant feeding artery is originated from the descending thoracic aorta (*arrow*) **(A)** and an aberrant aneurysm **(B)**.

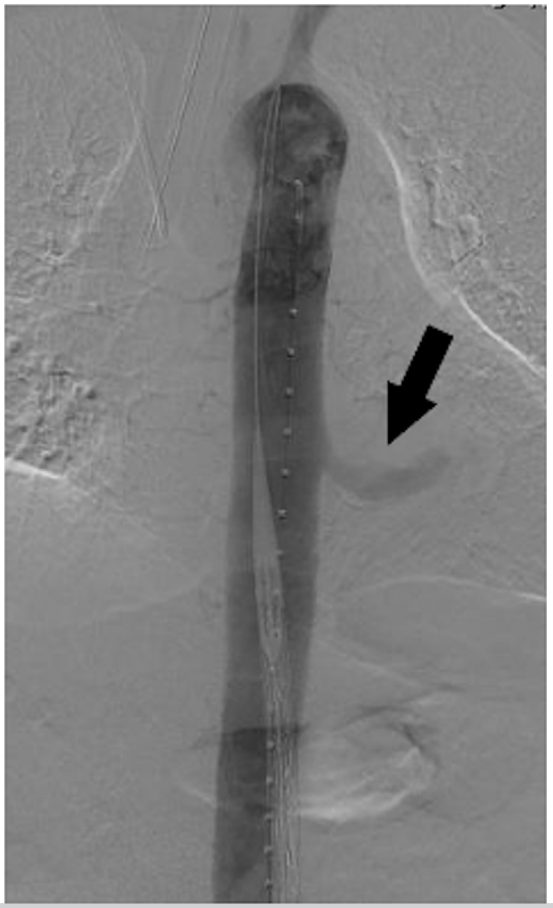

Fig 2. Angiogram reveals that an aberrant feeding artery is originated from the descending thoracic aorta.

ligation of the aberrant feeding artery and segmental lobectomy; isolated treatment of the aberrant feeding artery, including surgical ligations or division under thoracotomy or video-assisted thoracoscopic surgery; endovascular procedures, such as coil or plug embolization to the aberrant feeding artery or TEVAR to occlude it; and unifocalized anastomosis between the aberrant feeding artery and the pulmonary artery. The outcomes of both surgical repair and endovascular treatments for aberrant aneurysm with PS were good without major complications.[5-19] In these reports, surgical lobectomy was performed in eight cases; TEVAR and secondary lobectomy were chosen in five cases. Embolization was performed in two cases, one of which was by TEVAR. On the other hand, the rate of surgical repair complications of PS was reported to be 9.8% to 28%.[4,5] Embolization distal to the aneurysmal body should have been performed to attain complete thromboexclusion inside the aneurysmal body. However, coiling of all branches at the intended portion is difficult under arterial pressure. Therefore, in the present case, isolated treatment of the aberrant feeding artery was chosen based on these conditions: only the presence of chest discomfort, normal anatomy of both the pulmonary artery and bronchus to the left basal segment, and aneurysmal dilatation of the aberrant feeding artery. Also, TEVAR was thought to be the optimal treatment because of the short distance (3 mm in length) between the aneurysmal body and the orifice of the aberrant feeding artery originating from the descending thoracic aorta, which was considered to be inappropriate for surgical division/ligation of the target vessel or coil/plug embolization. Although TEVAR is a conventional and less invasive procedure, several complications related to TEVAR itself are of concern. Especially in the case of stent graft infection, a wider range of replacement of the diseased aorta along with the removal of the infected stent graft is usually crucial, which can be a much more demanding procedure, compared with the initial TEVAR. Because spinal cord ischemia is a different issue, a short device should be selected. Although there have been few reports with regard to the requirements of embolization to the vessels distal to the aneurysmal portion, Maekawa et al[2] reported that aneurysmal dilatation was not found after isolated ligation of the aberrant feeding artery ostium. In the present case, embolization distal to the aneurysmal

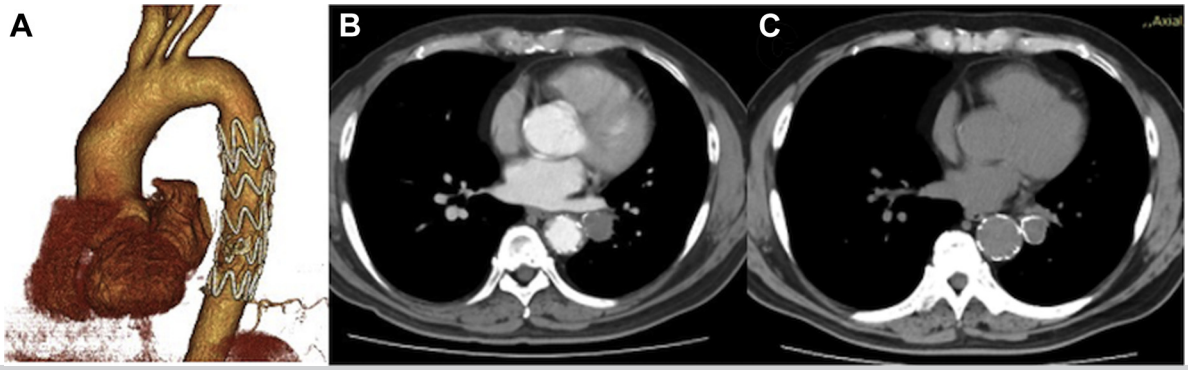

Fig 3. Postoperative computed tomography (CT) at 1 **(A and B)** and 6 **(C)** months after surgery reveals a significant shrinkage of the aberrant aneurysm.

body was not performed because multiple coiling materials were required to completely occlude all the branched vessels. This would complicate and encumber the procedure. To date, the present case demonstrates significant shrinkage of the aneurysmal body and thromboexclusion of the aberrant feeding artery. However, careful regular observation is necessary to monitor for inflammatory changes of the lung or aneurysmal reenlargement. We will follow up at 1-year regular intervals with the use of nonenhanced CT. If either situation occurs, segmental lobectomy via thoracotomy or video-assisted thoracoscopic surgery should be performed as a second stage procedure. To conclude, isolated TEVAR can be an optimal treatment option to correct anomalous systemic arterial supply to the left basal lung, in the case of normal anatomy of both the pulmonary artery and bronchus to the left basal segment, by simply occluding the aberrant feeding artery together with aneurysmal dilatation.

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Submitted Mar 7, 2021; accepted Jun 11, 2021.