

VASCULAR PLUG EMBOLIZATION OF TWO ARTERIES OF PULMONARY SEQUESTRATION PRIOR TO LOBECTOMY

LOBEKTOMİ ÖNCESİ PULMONER SEKESTRASYONUN VASKÜLER PLAK EMBOLİZASYONU

Ahmet Arif YALÇIN¹ (b), Serkan KAHRAMAN¹ (b), Murat AKKUŞ² (b), Aydın YILDIRIM¹ (b)

¹University of Health Sciences, Mehmet Akif Ersoy Thoracic and Cardiovascular Surgery Center, Training and Research Hospital, Department of Cardiology, Istanbul, Turkey

²University of Health Sciences, Mehmet Akif Ersoy Thoracic and Cardiovascular Surgery Center, Training and Research Hospital, Department of Thoracic Surgery, Istanbul, Turkey

ORCID IDs of the authors: A.A.Y. 0000-0002-9258-5389; S.K. 0000-0003-2796-0987; M.A. 0000-0002-3762-6328; A.Y 0000-0002-0766-1544

Cite this article as: Yalcin AA, Kahraman S, Akkus M, Yildirim A. Vascular plug embolization of two arteries of pulmonary sequestration prior to lobectomy. J Ist Faculty Med 2020;83(2):172-6. doi: 10.26650/IUITFD.2018.0021

ABSTRACT

A 40-year-old male patient was admitted to our emergency department with hemoptysis. A contrast-enhanced thoracic computed tomography demonstrated pulmonary sequestration supplied by two separate aberrant arteries. Due to the risk of excessive bleeding during elective surgery, transcatheter embolization of aberrant arteries was planned before the operation. A 10 mm Amplatzer Vascular Plug 2 was deployed to the proximal and a 6 mm Amplatzer Vascular Plug 1 was deployed to the distal artery via the right femoral artery. The procedure was terminated with successful vascular embolization of the two afore-mentioned arteries. One month after the embolization, the robotic left lower lobectomy was performed electively.

Keywords: Pulmonary sequestration, robotic lobectomy, vascular plugs

ÖZET

40 yaşında bilinen sistemik hastalık öyküsü olmayan hemoptizi şikayeti ile acil servisimize başvuran hastanın çekilen kontrastlı toraks BT'sinde sol alt akciğer parenkiminin torasik aortadan kaynaklı iki aberran arter ile kanlandığı saptanmıştır. Göğüs cerrahisi tarafından değerlendirilerek lobektomi planlanan hastanın peroperatif kanama riskini azaltmak için elektif cerrahi öncesi aberran arterlerin transkataterik kapatılması planlandı. Sağ femoral yolla yapılan anjiografi sonrası proksimal artere 10 mm Amplatzer Vasküler Plak 2, distal artere ise 6 mm Amplatzer Vasküler Plak 1 yerleştirilerek işlem başarı ile sonlandırıldı. Hastaya 1 ay sonra elektif olarak başarılı robotik lobektomi uygulandı. Bu hibrid yaklaşım nadir bir tedavi stratejisi olarak uygulanmıştır.

Anahtar Kelimeler: Pulmoner sekestrasyon, robotik lobektomi, vasküler plak

INTRODUCTION

Pulmonary sequestration is a rare congenital malformation that is characterized by the presence of lung tissue which does not have any communication with the normal bronchopulmonary tree and is supplied by one or more aberrant arteries (1). This condition is only likely to occur in 0.15-1.8% of the general population. There are two types of sequestration: intralober and extralober sequestration. Intralober sequestration that has its own separate pleura which is a more common type and generally presents in adults. Although it is well known that surgical treatment is recommended for symptomatic patients, transcatheter embolization is an effective treatment modality for patients at risk of excessive bleeding (2). We herein present a case of successful transcatheter vascular plug embolization of pulmonary sequestration patient with recurrent hemoptysis prior to robotic left lower lobectomy.

Corresponding author/İletişim kurulacak yazar: serkankahraman_86@outlook.com

Submitted/Başvuru: 06.10.2018 • Revision Requested/Revizyon Talebi: 18.09.2019 •

Last Revision Received/Son Revizyon: 25.09.2019 • Accepted/Kabul: 04.10.2019 • Published Online/Online Yayın: 06.03.2020 ©Telif Hakkı 2020 J Ist Faculty Med - Makale metnine jmed.istanbul.edu.tr web sayfasından ulaşılabilir.

©Teilf Hakki 2020 J ist Faculty Med - Makale metnine jmed.istanbul.edu.tr web sayfasindan ulaşılabilir ©Copyright 2020 by J lst Faculty Med - Available online at jmed.istanbul.edu.tr

CASE PRESENTATION

A 40-year-old male patient without any chronic disease was admitted to our emergency department with recurrent episodes of hemoptysis. He was a non-smoker and did not use any medication. His hemodynamic parameters including O_2 saturation were normal and also physical examination and routine laboratory investigations were unremarkable. While a chest radiograph did not have any significant pulmonary parenchymal or pleural abnormality,

thoracic computed tomography demonstrated pulmonary sequestration with increased parenchymal consolidation at the left lower pulmonary area (Figure 1A/B). A contrastenhanced thoracic computed tomography demonstrated left lower pulmonary parenchyma supplied by two separate aberrant arteries originating from lower thoracic aorta (Figure 2). The proximal aberrant artery (Figure 3A) was bigger than the distal aberrant artery (Figure 3B). The patient was evaluated by our division of thoracic surgery and a left lower lobectomy operation was



Figure 1: A-Chest radiograph of patient with pulmonary sequestration B-Thoracic computed tomography demonstrated pulmonary sequestration with increased parenchymal consolidation at the left lower pulmonary area (arrowhead).



Figure 2: Contrast-enhanced thoracic computed tomography demonstrated two separate aberrant arteries originating from lower thoracic aorta.



Figure 3: Contrast-enhanced thoracic computed tomography imaging of two aberrant arteries of pulmonary sequestration A- proximal artery B- distal artery.



Figure 4: Angiographic imaging of vascular plug embolization of aberrant arteries of pulmonary sequestration A-Angiographic imaging of proximal aberrant artery B-Angiographic imaging of distal aberrant artery C-Amplatzer Vascular Plug 2 implantation to proximal aberrant artery D-Cutting off the blood flow of the proximal artery E-Amplatzer Vascular Plug 1 implantation to distal aberrant artery F-Successful embolization of two aberrant arteries.

planned. Due to the risk of high bleeding during elective surgery, transcatheter embolization of aberrant arteries was approved before the operation by our cardiology and thoracic surgery team. The digital subtraction angiography was performed via the right common femoral artery using 7French (7F) femoral sheath. The two aberrant arteries were selectively cannulated with a Judkins Right (JR) 4-6F diagnostic catheter (Medtronic, New York, USA) and the angiographic images of theses arteries were obtained (Figure 4A/B). A 10 mm Amplatzer Vascular Plug 2 (AVP; St. Jude Medical, St. Paul, MN) was deployed to the proximal segment of the artery via 7F Destination sheat (Terumo Medical, Somerset, NJ) (Figure 4C). Then the blood flow of the artery was completely cut off (Figure 4D). After this, a 6 mm Amplatzer Vascular Plug 1 (AVP; St. Jude Medical, St. Paul, MN) was deployed in the distal one via Judkins Right (JR) 4-6F guiding catheter (Medtronic, New York, USA) (Figure 4E). The procedure was terminated with successful vascular embolization of the afore-mentioned two arteries (Figure 4F). The patient was discharged from hospital without any complication on the day after embolization. Although hemoptysis regressed comparatively, a surgical operation was planned by our cardiology and thoracic surgery team due to increased risk of recurrent inflammation at the sequestration area. It is known that interruption of the arterial supply of the sequestration is not enough to suppress the inflammation. One month after the embolization the robotic left lower lobectomy was performed electively.

DISCUSSION

Pulmonary sequestration is first described by Rektorzik as a congenital malformation of lung tissue which has its own arterial supply without any communication with the normal bronchopulmonary tree (3). Intralober sequestration having its own separate pleura is more common than the extralober type, and generally more than half of the cases of the intralober type present in later childhood or adulthood, while extralober is generally diagnosed in infants. Intralober sequestration is generally located in the lower lobes of lung tissue (98%) and arterial supply often arises from the lower thoracic aorta (4). 2625 patients are evaluated by Young et al. and the most common symptoms are detected as cough, sputum production, fever, hemoptysis and chest pain. 13% of patients presented without any symptom with incidental imaging findings (5). While the digital subtraction angiography is the gold standard procedure for detection of abnormal arterial supply, contrast-enhanced thoracic computed tomography and magnetic resonance imaging are alternative options.

Surgical resection of pulmonary sequestration is the most commonly accepted treatment procedure. However, some patients, having more than one aberrant artery or having arteries arising from the posterolateral area of the aorta, are under high risk of surgical bleeding, such as our patient. Spontaneous regression of pulmonary sequestration due to progressive fibrosis of the dysplastic lung tissue is seen to be difficult in these patients. The other possible mechanism of induced hemorrhage risk is based on friable arterial supply of sequestration due to inflammation from repeated infection. Embolization of the aberrant arteries mimics this mechanism by occluding the arterial supply and it has been demonstrated that embolization can reduce the risk of intraoperative hemorrhage (6). Moreover, embolization can be an alternative treatment strategy of surgical lobectomy or it can be performed prior to surgery in patients under high risk of intraoperative hemorrhage such as our patient. According to the mentioned strategy, our patient was treated successfully with vascular plug embolization of two aberrant arteries of the sequestration to reduce the risk of bleeding during surgery. While there is no data in the literature regarding the optimal timing of the surgical procedure after embolization, one month after embolization, the surgical robotic lobectomy, having a low perioperative hemorrhage risk, was performed electively on the decision of our cardiology and thoracic surgery team. For a month after the embolization had been carried out antiplatelet monotherapy with was given to reduce thromboembolic occurrence. Thus, a surgical operation was performed one month after the procedure. This hybrid procedure is quite a rare option in the treatment of pulmonary sequestration due to the high perioperative risk of bleeding due to more than one aberrant artery. It has been reported that vascular embolization has some complications including femoral arterial access site thrombosis and transient limb ischemia, non-target embolization of the pulmonary arteries and aorta, transient hypertension, pain, and fever (2,5). Adamkiewicz arterial occlusion is one of the most serious complications of this procedure. Some of these complications can give rise to increased morbidity and mortality rates. However, the complication rates of vascular plug embolization are low in an experienced center.

CONCLUSION

Pulmonary sequestration is a rare congenital malformation of lung tissue with aberrant arterial supply. Although surgical lobectomy is the gold standard treatment, vascular embolization is the common accepted strategy that is used instead of or prior to surgery in selected patients. Vascular plug embolization of two aberrant arteries of the sequestration in the same procedure prior to surgical robotic lobectomy is a rare type of treatment. It can enable robotic surgery to reduce the risk of intraoperative hemorrhage. Furthermore, studies on a large scale are needed to evaluate the long term outcome of vascular plug embolization.

Etik Komite Onayı: Etik komite onayı alınmamıştır.

Bilgilendirilmiş Onam: Katılımcılardan bilgilendirilmiş onam alınmıştır.

Hakem Değerlendirmesi: Dış bağımsız.

Yazar Katkıları: Çalışma Konsepti/Tasarım- A.A.A, S.K., M.A., A.A.; Veri Toplama- A.A.A, S.K.; Veri Analizi/Yorumlama- B.K, N.B.; Yazı Taslağı- A.A.A, S.K.; İçeriğin Eleştirel İncelemesi-A.A.A, S.K., M.A., A.A.; Son Onay ve Sorumluluk- A.A.A, S.K., M.A., A.A.; Malzeme ve Teknik Destek- A.A.Y.; Süpervizyon-A.A.A, S.K., M.A., A.A.

Finansal Destek: Yazarlar finansal destek beyan etmemişlerdir.

Çıkar Çatışması: Yazarlar çıkar çatışması beyan etmemişlerdir.

Ethics Committee Approval: This study was not approved by an ethical committee.

Informed Consent: Written consent was obtained from the participants.

Peer Review: Externally peer-reviewed.

Author Contributions: Conception/Design of Study- A.A.A, S.K., M.A., A.A.; Data Acquisition- A.A.A, S.K.; Data Analysis/Interpretation- A.A.A, S.K., M.A., A.A.; Drafting Manuscript- A.A.A, S.K.; Critical Revision of Manuscript- B A.A.A, S.K., M.A., A.A.; Final Approval and Accountability- A.A.A, S.K., M.A., A.A.; Technical or Material Support- A.A.Y.; Supervision- A.A.A, S.K., M.A., A.A.

Conflict of Interest: Authors declared no conflict of interest.

Financial Disclosure: Authors declared no financial support.

REFERENCES

 Yucel O, Gurkok S, Gozubuyuk A, Caylak H, Sapmaz E, Kavakli K, et al. Diagnosis and surgical treatment of pulmonary sequestration. Thorac Cardiovasc Surg 2008;56(3):154-57. [CrossRef]

- Ganeshan A. Freedman J, Hoey ET, Steyn R, Henderson J, Crowe PM. Transcatheter coil embolization: a novel definitive treatment option for intralobar pulmonary sequestration. Heart Lung Circ 2010;19(9):561-5. [CrossRef]
- Felker RE, Tonkin IL. Imaging of pulmonary sequestration. AJR Am J Roentgenol 1990;154(2):241-9. [CrossRef]
- Savic B, Birtel FJ, Tholen W, Funke HD, Knoche R. Lung sequestration: Report of seven cases and review of 540 published cases. Thorax 1979;34(1):96-101. [CrossRef]
- Yong W, Fan L. Pulmonary sequestration: a retrospective analysis of 2625 cases in China. Eur J Cardiothorac Surg 2011;40(1):39-42. [CrossRef]
- Tokel K, Boyvat F, Varan B. Coil embolization of pulmonary sequestration in two infants; a safe alternative to surgery. AJR Am J Roentgenol 2000;175(4):993-5. [CrossRef]