© 2024 The Authors. Published by Elsevier Inc. on behalf of The Society of Thoracic Surgeons.
This is an open access article under the CC BY-NC-ND license
(http://creativecommons.org/licenses/by-nc-nd/4.0/).
2772-9931
https://doi.org/10.1016/j.atssr.2023.12.011

Lung: Case Report

Successful Treatment of Multiple Systemic Artery–to–Pulmonary Artery Fistulas

Taiki Ryo, MD,[1] Kikuo Shigemitsu, MD,[1] Shunsuke Mori, MD, PhD,[1] Atsushi Enomoto, MD, PhD,[2] and Toyofumi Fengshi Chen-Yoshikawa, MD, PhD[3]

Systemic artery–to–pulmonary artery fistula (SAPAF) is an uncommon abnormal vascular connection between systemic and pulmonary arteries. SAPAF with 3 or more inflow vessels has rarely been reported. The definitive diagnosis is made by selective arterial angiography, but 3-dimensional computed tomography angiography is useful for delineating abnormal vessels. Embolization is currently performed as a less invasive treatment option, but surgical treatment remains an essential treatment option for preventing recurrence, especially in cases with many abnormal vessels. Herein, we report a case of successful treatment of SAPAF by sparing the lung parenchyma through abnormal inflow vessel dissection and peripheral lung abnormal tissue resection.

(Ann Thorac Surg Short Reports 2024;2:172-174)
© 2024 The Authors. Published by Elsevier Inc. on behalf of The Society of Thoracic Surgeons. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

Systemic artery-to-pulmonary artery fistula (SAPAF) is an uncommon abnormal vascular connection between systemic and pulmonary arteries.[1] Most cases are consequences of trauma, surgery, infection, or inflammatory disease, but congenital cases account for approximately 15%.[2] Examples of inflow vessels include bronchial arteries, internal thoracic arteries, and coronary arteries.[3-5] SAPAF with 3 or more inflow vessels has rarely been reported.[3] The definitive diagnosis is made by selective arterial angiography, but 3-dimensional computed tomography angiography (3D-CTA) is useful for delineating abnormal vessels. Most patients are asymptomatic, but several authors stated that treatment intervention is recommended because of the risk of further complications, such as infection, bleeding, pulmonary hypertension, and heart failure.[4] Embolization is currently performed as a less invasive treatment option; however, surgical treatment remains an essential option for treating SAPAF and preventing its recurrence, especially in cases with many abnormal vessels.[5]

A 67-year-old woman demonstrated abnormal shadows on the chest radiograph during a physical examination. She was asymptomatic and had no history of chest surgery, trauma, pneumonia, and other lung diseases. Examination revealed no evidence of wheezing or cardiac murmurs, and blood and respiratory function test results were normal. Abnormal vessels of celiac artery origin, left internal thoracic artery, intercostal artery flowing into the left lingual segment, and aneurysmally dilated pulmonary artery were seen on 3D-CTA (Figure 1A). Because of these various abnormal vessels and the tortuosity, surgical treatment was thought to be a better treatment option than embolization in terms of nonrecurrence.

Surgical treatment was performed through mini-thoracotomy by thoracoscopy. A markedly tortuous sixth intercostal artery was seen when the chest was opened at the sixth intercostal space. The tip of the lingual segment was adherent to the pericardial fat, and abnormal vessels from the left internal thoracic and celiac arteries could be seen flowing into it (Figure 2A). Palpation over these areas revealed a prominent systolic thrill. The sixth intercostal artery was ligated and dissected. The left internal thoracic and celiac arteries were ligated and dissected with an automatic suture machine (Figure 2B). In addition, the abnormal vessels derived from the internal thoracic artery, which were partially running on the pericardium and flowing into the mediastinal aspect of the lingual segment, were treated with a vascular sealing device. Then, the tip of the lingual segment collapsed without any pulsation. This abnormal peripheral lung was eventually resected with an automatic suture machine.

The patient had an uneventful postoperative course, and she was discharged on postoperative day 6. Histologic examination of the resected lung revealed abnormal vessels with an extremely heterogeneous thickness of tunica media elastic fibers and a sparse density of outer membrane connective tissue (Figure 3). Contrast-enhanced computed tomography (CT) at the 6-month postoperative follow-up revealed improvement

Accepted for publication Dec 18, 2023.

[1]Department of Thoracic Surgery, Ogaki Municipal Hospital, Gifu, Japan; [2]Department of Pathology, Graduate School of Medicine, Nagoya University, Aichi, Japan; and [3]Department of Thoracic Surgery, Graduate School of Medicine, Nagoya University, Aichi, Japan

Address correspondence to Dr Ryo, Department of Thoracic Surgery, Ogaki Municipal Hospital, 4-86 Minaminokawa-chou, Ogaki-shi, Gifu, 503-0807, Japan; email: taikiryo0715@gmail.com.

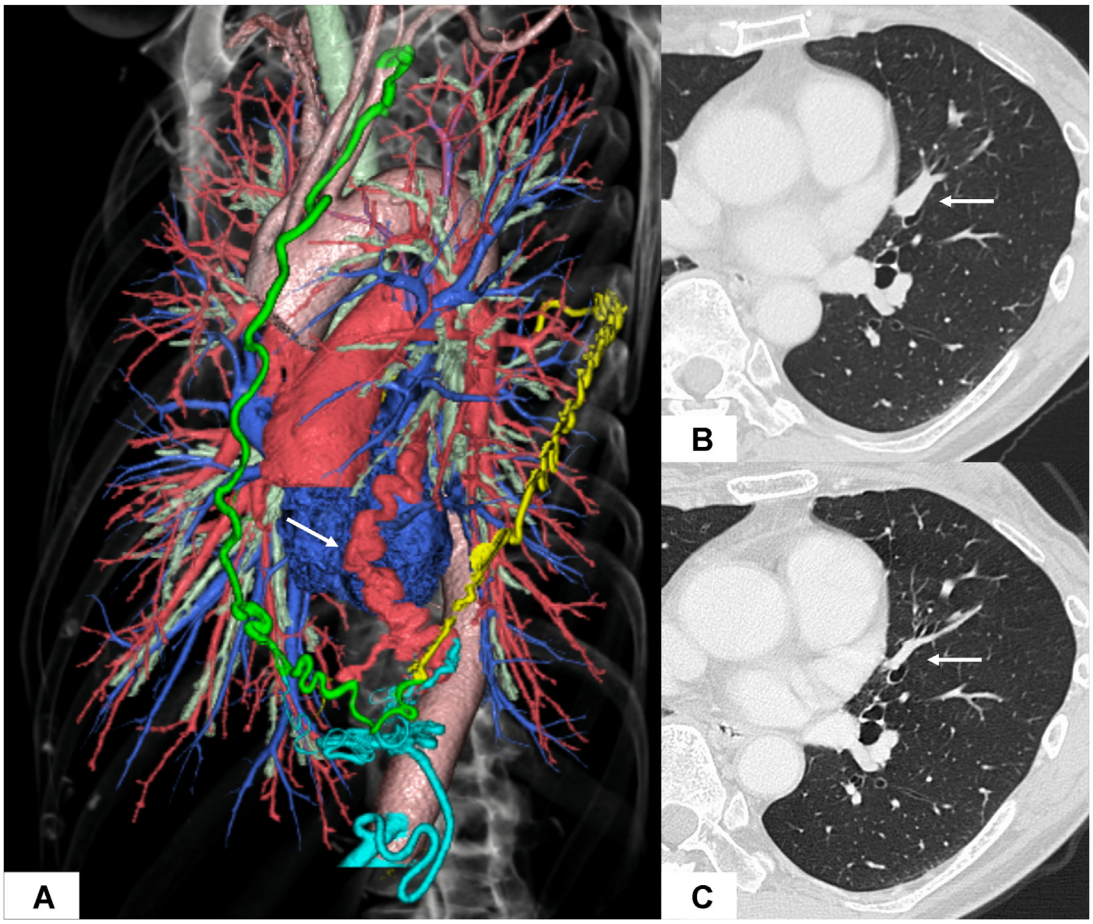

FIGURE 1 Multiple systemic artery–to–pulmonary artery fistulas on 3-dimensional computed tomography angiography. (A) Abnormal vessels of celiac artery origin (blue), left internal thoracic artery (green), and intercostal artery (yellow) flowing into an aneurysmally dilated pulmonary artery (arrow). Contrast-enhanced computed tomography (B) preoperatively and (C) postoperatively. Pulmonary artery dilation was remarkably improved (arrows).

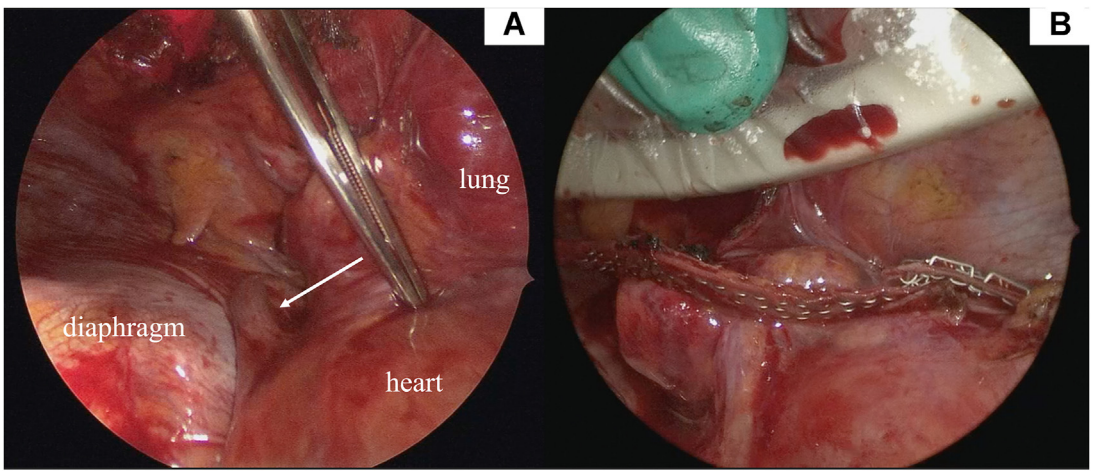

FIGURE 2 Intraoperative findings. (A) Abnormal vessels (arrow) were seen flowing into the pericardial fat. (B) They were ligated and dissected with an automatic suture machine.

Ann Thorac Surg Short Reports
2024;2:172-174

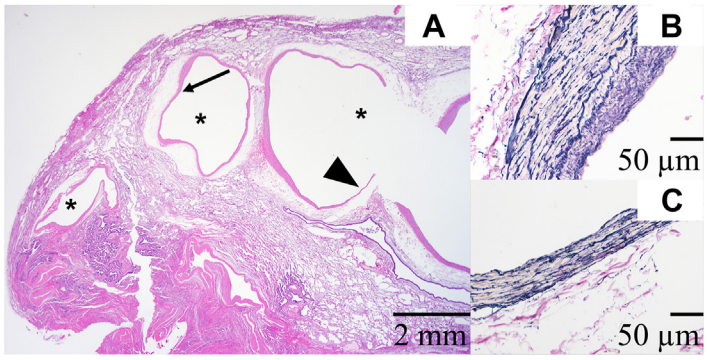

FIGURE 3 Histologic examination of the resected lung. (A) Hematoxylin and eosin–stained abnormal vessels (asterisk) in the resected lung. (B, C) Elastica van Gieson stain. Abnormal vessels with an extremely uneven thickness of tunica media elastic fibers and sparse connective tissue density of the outer membrane were observed (B, magnification of arrow shown in A; C, magnification of arrowhead shown in A).

of the pulmonary artery, which was tortuous and dilated preoperatively (Figures 1B, 1C). Finally, the patient was diagnosed with SAPAF on the basis of these findings and has been disease free for 2 years after the operation.

COMMENT

SAPAF with inflow vessels from 3 or more different systemic arteries is extremely rare and has hardly been reported.[3] Three-dimensional image reconstruction has been widely performed in thoracic surgery,[6] and 3D-CTA visualized abnormal vascular anastomoses with minimal invasiveness in this patient. Selective arterial angiography was not performed because of the obvious abnormal vessels. However, performing selective arteriography might be better in terms of preoperative definitive diagnosis because CT cannot show hemodynamics such as regurgitation into the pulmonary artery or shunt rate and minor abnormal feeders. Notably, postoperative contrast-enhanced CT revealed improvement of the tortuous and dilated pulmonary artery compared with preoperative CT, which is important evidence of return flow to the pulmonary artery. The patient was finally diagnosed with SAPAF on the basis of the histologic findings in addition to this CT finding.

In terms of treatment, embolization has recently been performed as a less invasive treatment because it causes minor trauma and does not require general anesthesia.[3,4] Surgical intervention remains an important treatment option to ensure a single treatment and to prevent a recurrence, especially in patients with many abnormal vessels, such as in this case.[5] Embolization has a potential risk of recurrence, and the presence of SAPAF is reported as an independent risk factor predicting early recurrence of hemoptysis after embolization.[7] Various surgical treatments, such as lobectomy, segmentectomy, and pneumonectomy, have been reported,[8] but only abnormal inflow vessel dissection and partial peripheral lung resection could spare the normal lung parenchyma in this patient.

In conclusion, 3D-CTA was useful in diagnosing asymptomatic SAPAF. We successfully treated the patient by sparing the lung parenchyma by abnormal inflow vessel dissection and peripheral lung abnormal tissue resection.

The authors wish to thank Enago (www.enago.jp) for the English language review. This study was approved by the institutional review board of Ogaki Municipal Hospital (20230427-20-c; April 28, 2023).

FUNDING SOURCES
The authors have no funding sources to disclose.

DISCLOSURES
The authors have no conflicts of interest to disclose.

PATIENT CONSENT
Written informed consent was obtained from the patient.

REFERENCES

1. Burchell HB, Clagett OT. The clinical syndrome associated with pulmonary arteriovenous fistulas, including a case report of a surgical cure. *Am Heart J*. 1947;34:151-162.

2. Abdul Jabbar A, Patel A, Marzlin N, et al. Internal mammary artery–to–pulmonary vasculature fistula: systematic review of case reports. *Vasc Med*. 2017;22:426-431.

3. Lee JK, Park JH, Kim J, et al. Embolization of multiple systemic artery to pulmonary artery fistula with recurrent hemoptysis. *Tuberc Respir Dis (Seoul)*. 2013;75:120-124.

4. Yamada Y, Imamura H, Amamoto Y, Ochi M, Nagano K, Ito M. Congenital internal mammary artery–to–pulmonary artery fistulas: a case report. *Heart Vessels*. 1987;3:47-49.

5. Matsudo K, Haratake N, Ono Y, et al. Multiple systemic artery to pulmonary vessel fistulas (SAPVFs) completely resected by video-assisted thoracoscopic surgery: a case report. *Surg Case Rep*. 2022;8:184.

6. Chen-Yoshikawa TF, Date H. Update on three-dimensional image reconstruction for preoperative simulation in thoracic surgery. *J Thorac Dis*. 2016;8:S295-S301.

7. Zhang CJ, Jiang FM, Zuo ZJ, et al. Clinical characteristics and postoperative outcomes of systemic artery–to–pulmonary vessel fistula in hemoptysis patients. *Eur Radiol*. 2022;32:4304-4313.

8. Shang HW, Sun SB, Ma GY, Mei XM, Li C, Yang K. A ruptured pulmonary arteriovenous fistula after laparoscopic operation. *Chin J Traumatol*. 2017;20:359-361.