

Thoracic Endovascular Aortic Repair With Subclavian Revascularization for Symptomatic Nonaneurysmal Aberrant Right Subclavian Artery

Yuki Nakamura, MD, PhD¹; Shusuke Imaoka, MD²; Takuya Yamakura, MD¹; Taro Yamasumi, MD¹; Haruhiko Kondoh, MD, PhD¹

¹Department of Cardiovascular Surgery, Japan Organization of Occupational Health and Safety, Osaka Rosai Hospital, Sakai, Osaka, Japan

²Department of Cardiovascular Surgery, Osaka University Graduate School of Medicine, Osaka, Japan

Aberrant right subclavian artery is a common aortic arch anomaly that can cause dysphagia as a result of compression by the aberrant artery. For patients with an aneurysm associated with an aberrant right subclavian artery, surgical or endovascular intervention is a well-described treatment. However, for patients with a nonaneurysmal aberrant right subclavian artery, treatment with thoracic endovascular aortic repair has been limited. We describe the use of thoracic endovascular aortic repair and subclavian revascularization to treat esophageal stricture in a patient with a symptomatic nonaneurysmal aberrant right subclavian artery. The patient's dysphagia was successfully relieved after the operation. (Tex Heart Inst J 2022;49(4):e207489)

Citation:

Nakamura Y, Imaoka S, Yamakura T, Yamasumi T, Kondoh H. Thoracic endovascular aortic repair with subclavian revascularization for symptomatic nonaneurysmal aberrant right subclavian artery. *Tex Heart Inst J* 2022;49(4):e207489. doi: [10.14503/THIJ-20-7489](https://doi.org/10.14503/THIJ-20-7489)

Key words:

Aorta, thoracic; deglutition disorders; dysphagia; endovascular procedures/methods; esophageal stenosis; subclavian artery/abnormalities/diagnostic imaging/surgery; vascular malformations

Corresponding author:

Haruhiko Kondoh, MD, Department of Cardiovascular Surgery, Japan Organization of Occupational Health and Safety, Osaka Rosai Hospital, 1179-3, Nagasone-cho, Sakai 591-8025, Japan

E-mail:

haruhk@wa3.so-net.ne.jp

© 2022 by the Texas Heart[®] Institute, Houston

A aberrant right subclavian artery (ARSA) is a common type of aortic arch anomaly, occurring in 0.5% of the population.¹ Although most patients with ARSA are asymptomatic, dysphagia (ie, dysphagia lusoria) or respiratory symptoms caused by ARSA compression have been observed in 7% to 10% of adult patients with this anomaly.²

In approximately 60% of ARSA cases, the aneurysmal origin is due to noninvolution of the right dorsal aorta (Kommerell diverticulum).³ Patients in these cases are usually treated through surgical or endovascular intervention. For patients with nonaneurysmal ARSA, treatment with thoracic endovascular aortic repair (TEVAR) has been reported in only 2 cases to date,⁴ and the indications and optimal treatment strategies remain controversial. We describe the successful use of TEVAR and subclavian revascularization for esophageal stricture in a patient with symptomatic nonaneurysmal ARSA.

Case Report

An otherwise healthy 60-year-old man presented at our hospital with dysphagia, weight loss, and the ability to swallow only liquid food. A contrast-enhanced computed tomographic (CT) angiogram indicated nonaneurysmal ARSA. The right subclavian artery was found to arise from the aortic arch as a fourth branch, coursing posterior to the esophagus (Fig. 1). An upper gastrointestinal endoscopic examination showed no notable abnormalities in the pharynx or esophagus, including esophageal stenosis; however, an esophagogram revealed an esophageal defect (Fig. 2). Although we suspected that ARSA was the most likely cause of dysphagia, it was not clear whether ARSA had caused the esophageal compression that subsequently led to dysphagia. Therefore, we performed an endovascular intervention to improve the patient's symptoms, with plans to perform open surgical ligation of the ARSA if they did not.

The patient was taken to a hybrid operating room, and general anesthesia was administered. A bilateral supraclavicular incision was made to expose the bilateral subclavian arteries and the bilateral carotid arteries to prepare for eventual bilateral

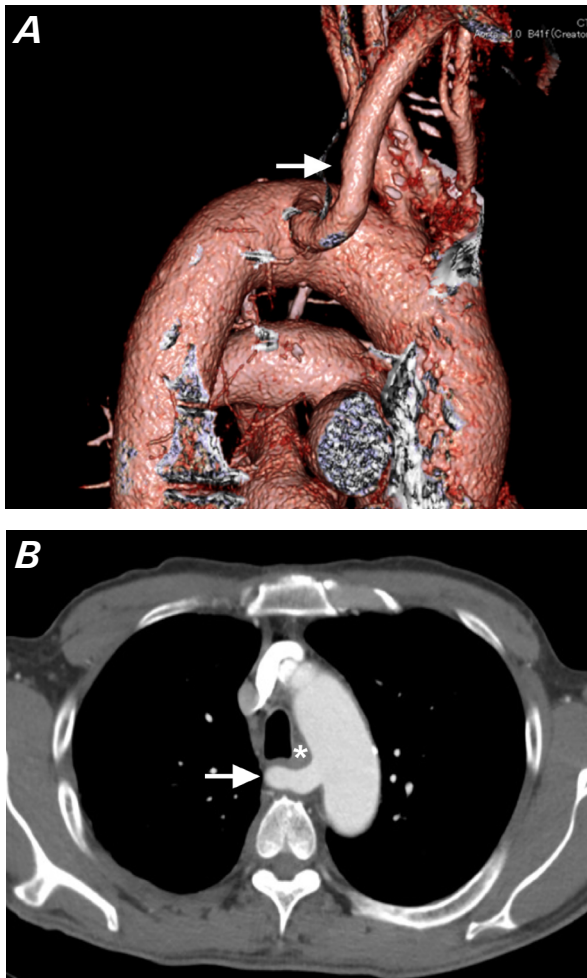


Fig. 1 Preoperative computed tomographic angiograms in **A)** 3-dimensional reconstruction and **B)** axial views show a nonaneurysmal aberrant right subclavian artery (arrow). In **B**, note the nonaneurysmal artery coursing posterior to the esophagus (asterisk).

carotid-subclavian artery bypass. Simultaneously, a right inguinal incision was made to expose the right common femoral artery. The patient was systemically heparinized, and methylprednisolone was administered. A bilateral carotid-subclavian artery bypass was performed in standard fashion with use of an 8-mm expanded polytetrafluoroethylene graft. A 5F sheath was then inserted in the bilateral subclavian arteries proximal to the anastomosis sites. Balloon-tip catheters were placed at a site in the left subclavian artery just proximal to the left vertebral artery and at the origin of the right subclavian artery through the 5F sheath. A 6F sheath was inserted into the right common femoral artery and then scaled up to a 22F sheath. A 34-mm × 15-cm TAG stent-graft (W.L. Gore & Associates) was inserted through the 22F sheath and deployed just distal to the origin of the left carotid artery. After the stent-graft was attached with use of a Tri-Lobe balloon catheter (W.L. Gore & Associates), the left subclavian artery was occluded by plac-

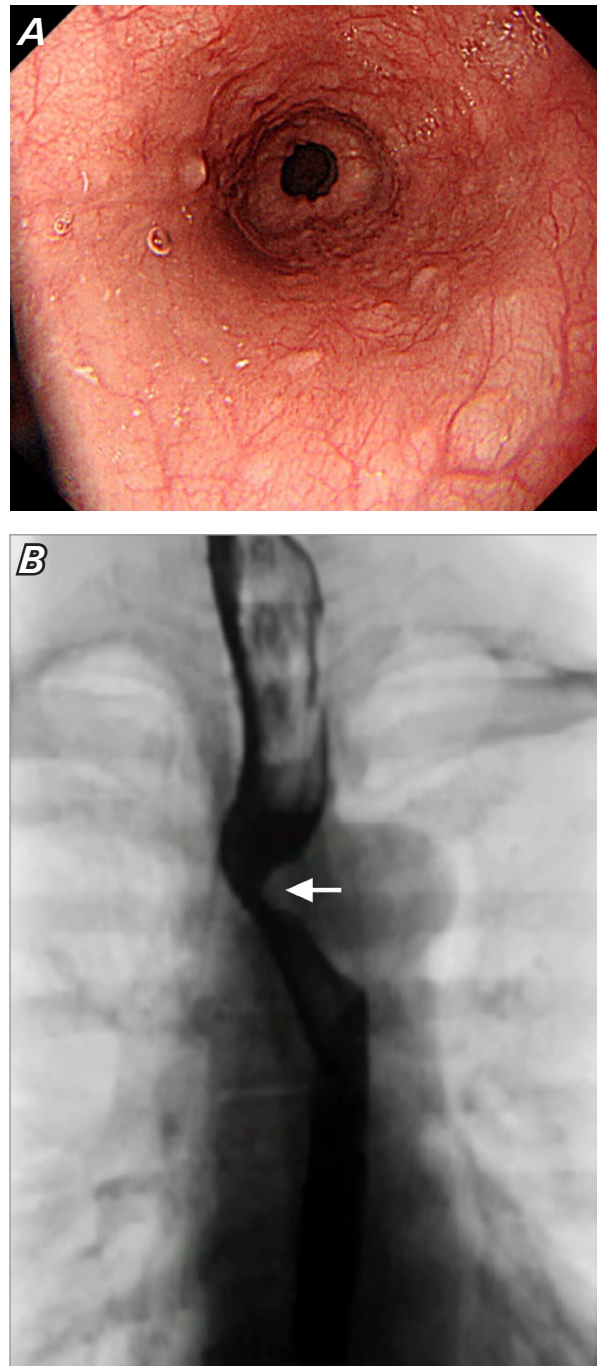


Fig. 2 Preoperatively, **A)** an upper gastrointestinal endoscopic image shows no significant stenosis, but **B)** an esophagogram shows an esophageal stricture (arrow).

ing an Interlock-35 coil (Boston Scientific) at the origin of the left subclavian artery. Finally, the right subclavian artery was occluded by placing an Amplatzer Vascular Plug II (Abbott Laboratories) at the distal end of the right subclavian artery adjacent to the esophagus. After completion, an angiogram showed slight leakage into the ARSA, which was suspected to be diminished by the occlusion of the right subclavian artery with the Amplatzer plug.

The patient's postoperative course was uneventful. After the operation, his dysphagia improved, and he resumed a regular diet on postoperative day 13. A postoperative esophagogram showed no substantial esophageal stenosis. A postoperative CT angiogram showed a minor proximal endoleak into the ARSA, but no other abnormality (Fig. 3). Because the patient's symptoms improved, no further intervention was performed for the endoleak. The patient was discharged on postoperative day 17. During the 4-year follow-up period, the patient's condition remained stable with no recurrence of symptoms. A follow-up CT angiogram showed gradual reduction of the endoleak into the ARSA.

Discussion

Treating our patient's symptomatic nonaneurysmal ARSA with TEVAR and revascularization markedly improved his dysphagia. Patients with ARSA are usually asymptomatic, but the most common and well-recognized manifestation of ARSA in adults is dysphagia lusoria.⁵ Dysphagia related to ARSA typically develops during the fourth or fifth decade of life and can often be explained by the presence of an ARSA-associated aneurysm. However, in the absence of an aneurysm, the reason for the late-onset dysphagia is unclear.⁶ Several possible mechanisms have been suggested, such as artery elongation and aging-related atherosclerosis.⁴ Given our patient's nonaneurysmal ARSA, these factors may have contributed to his dysphagia.

Aberrant right subclavian artery is easy to diagnose with use of computed tomography. However, findings indicating esophageal compression, which can cause dysphagia, are not sufficient for an ARSA diagnosis, especially in the absence of aneurysm formation. For this reason, we also examined our patient endoscopically and esophagographically. Whereas endoscopy showed no obvious stenosis of the esophagus, esophagography revealed stenosis and stagnation of the contrast agent just above the stenotic lesion. Therefore, despite the absence of an aneurysm, we considered ARSA compression a highly likely cause of our patient's dysphagia. We think that multiple imaging modalities should be used to confirm the existence of esophageal compression resulting from a nonaneurysmal ARSA.

Patients with an ARSA who have no symptoms or no associated aneurysm can be monitored without intervention; however, treatment should be considered if symptoms develop, even in the absence of an aneurysm.⁴ The first-line treatment option for symptomatic ARSA is surgery involving a left thoracotomy with resection of the ARSA from its origin to the right border of the esophagus, as described in the report of the first successful repair by Gross.⁷ More recently, case reports of 2 adolescent patients (13 and 16 years old) have shown that surgical division of nonaneurysmal ARSA is important



Fig. 3 Postoperative angiogram shows patent bilateral carotid-subclavian artery bypasses (arrows) and exclusion of the nonaneurysmal aberrant right subclavian artery with use of an Amplatzer vascular plug II (asterisk and bracket).

for treating dysphagia.^{8,9} However, avoiding a thoracotomy may reduce the rate of morbidity associated with open surgery, and such an approach may be especially suitable for patients with nonaneurysmal ARSA. Tinelli and colleagues¹⁰ reported good midterm outcomes in patients treated with a hybrid procedure involving TEVAR with graft exclusion of the aneurysmal ARSA and revascularization. Although this hybrid approach is considered feasible and safe in patients with aneurysmal ARSA, reports of its use in patients with nonaneurysmal ARSA are few.⁴ Leon and associates¹¹ reported the successful ligation of a symptomatic nonaneurysmal ARSA with subclavian-to-carotid transposition of the ARSA followed by endovascular occlusion of the stump with use of an Amplatzer vascular plug. Although minimally invasive, this alternative hybrid procedure is also associated with the potential risk that the endovascular device occluding the proximal site of the ARSA may migrate, erode the vessel wall, and cause an esophageal fistula or dysphagia by direct compression.¹² Kedora and colleagues¹³ also described occlusion of the proximal site of an aneurysmal ARSA with a plug. In their case, the plug was positioned in the subclavian arterial segment overlying the esophagus; however, dysphagia persisted because of compression caused by the plug itself. Furthermore, the close proximity of the subclavian artery

to the esophagus potentially increases the risk of a subclavian artery–esophageal fistula, which often has fatal consequences.¹⁴ Thus, pure occlusion of the proximal site of the ARSA with a plug may lead to an unsuccessful outcome in the postoperative period. We think, however, that this complication may be more likely to occur in patients with a nonaneurysmal ARSA than in those with an aneurysmal ARSA because the vascular lumen is narrower. To avoid having the plug directly compress the esophagus, the endovascular device should be placed on the distal end of the right aberrant artery, away from the esophagus.

Jalaie and associates⁴ reported outcomes in 2 patients (53 and 54 years old) whose nonaneurysmal ARSAs were treated with a hybrid procedure comprising TEVAR and revascularization of the ARSA. In both cases, successful relief of dysphagia was noted after the operation. However, midterm outcomes were not described for these patients and therefore cannot be compared with the good midterm outcomes seen in patients with aneurysmal ARSA who underwent TEVAR with revascularization.¹⁰ In the patient treated by Leon and colleagues¹¹ with a hybrid procedure involving nonaneurysmal ARSA ligation and subclavian-to-carotid transposition followed by endovascular closure of the ARSA origin, follow-up was performed only during the first 6 months after the operation. In our patient with symptomatic nonaneurysmal ARSA, dysphagia had not reoccurred 4 years after he underwent TEVAR with subclavian revascularization for esophageal stricture. Therefore, we think that minimally invasive TEVAR with revascularization of the ARSA may produce an effective, durable outcome regardless of whether the ARSA is aneurysmal or nonaneurysmal.

Conclusion

Minimally invasive TEVAR with revascularization is a feasible therapeutic option for patients with symptomatic nonaneurysmal ARSA and may result in good early and midterm outcomes. However, further investigation in a larger sample population and for a longer follow-up period is warranted to determine the best operative technique in these patients.

Published: 30 August 2022

Conflict of Interest Disclosures: None

Funding/Support: None

References

1. Stewart JR, Kincaid OW, Edwards JE. An atlas of vascular rings and related malformations of the aortic arch system. Springfield: Charles C Thomas; 1964. p.52-75, 98-123.
2. Delap TG, Jones SE, Johnson DR. Aneurysm of an aberrant right subclavian artery presenting as dysphagia lusoria. *Ann Otol Rhinol Laryngol* 2000;109(2):231-4.
3. Brown DL, Chapman WC, Edwards WH, Coltharp WH, Stoney WS. Dysphagia lusoria: aberrant right subclavian artery with a Kommerell's diverticulum. *Am Surg* 1993;59(9):582-6.
4. Jalaie H, Grommes J, Sailer A, Greiner A, Binnebösel M, Kalder J, et al. Treatment of symptomatic aberrant subclavian arteries. *Eur J Vasc Endovasc Surg* 2014;48(5):521-6.
5. Bayford D. An account of a singular case of obstructed deglutition. *Mem Med Soc Lond* 1794;2:275-86.
6. Klinkhamer AC. Aberrant right subclavian artery: clinical and roentgenologic aspects. *Am J Roentgenol Radium Ther Nucl Med* 1966;97(2):438-46.
7. Gross RE. Surgical treatment for dysphagia lusoria. *Ann Surg* 1946;124:532-4.
8. Darwazah AK, Eida M, Khalil RA, Ismail H, Hanbali N. Non-aneurysmal aberrant right subclavian artery causing dysphagia in a young girl: challenges encountered using supraclavicular approach. *J Cardiothorac Surg* 2015;10:92.
9. Thompson JL, Burkhart HM. Translocation of an aberrant right subclavian artery with resolution of dysphagia lusoria. *Ann Thorac Surg* 2016;102(1):e65-7.
10. Tinelli G, Ferrer C, Giudice R, Ferraresi M, Pogany G, Cao P, et al. Long-term results of hybrid repair techniques for Kommerell's diverticulum. *J Vasc Surg* 2020;72(4):1213-21.
11. Leon M, Garibaldi M, Virgen F, Ramirez-Cerda C, Cohen-Mussali S. Hybrid treatment of aberrant right subclavian artery causing dysphagia lusoria by subclavian to carotid transposition and endovascular plug. *Vasc Specialist Int* 2020;36(4):258-62.
12. Soo Hoo AJ, Rokkas CK, Rossi PJ. Migration of endovascular plug in hybrid repair of dysphagia lusoria. *J Vasc Surg Cases Innov Tech* 2018;4(2):140-3.
13. Kedora J, Grimsley B, Pearl G. Endovascular treatment of an aberrant right subclavian artery aneurysm with use of the Zenith iliac plug. *Proc (Bayl Univ Med Cent)* 2009;22(2):144-5.
14. Eggebrecht H, Mehta RH, Dechene A, Tsagakis K, Kühl H, Huptas S, et al. Aorto-esophageal fistula after thoracic aortic stent-graft placement: a rare but catastrophic complication of a novel emerging technique. *JACC Cardiovasc Interv* 2009;2(6):570-76.