



Anomalous Origin of the Left Anterior Descending Coronary Artery in an Adult

Keisuke Shibagaki¹ Chikara Shiiku² Hiroyuki Kamiya¹ Yoichi Kikuchi²

¹Department of Cardiac Surgery, Asahikawa Medical University, Asahikawa, Japan

²Department of cardiovascular surgery, National Obihiro Hospital, Obihiro, Hokkaido, Japan

Address for correspondence Hiroyuki Kamiya, MD, PhD, Department of Cardiac Surgery, Asahikawa Medical University, Midorigaoka Higashi 2-1-1, Asahikawa 078-8510, Japan (e-mail: kamiya@asahikawa-med.ac.jp).

Thorac Cardiovasc Surg Rep 2021;10:e9–e11.

Abstract

Keywords

- ▶ coronary anomalies
- ▶ ALADPA
- ▶ coronary artery bypass grafting

An anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) is a rare congenital heart disease. Among the variants, an anomalous origin of the left anterior descending coronary artery from the pulmonary artery (ALADPA) is extremely rare. Here, we report a case of ALADPA in an adult that was treated with coronary artery bypass grafting using the left internal thoracic artery.

Introduction

An anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) is a rare congenital heart disease. Among the variants, an anomalous origin of the left anterior descending coronary artery from the pulmonary artery (ALADPA) is extremely rare,¹ and there have been only a few reports of ALADPA in adults. We report a case of ALADPA in an adult that was treated with coronary artery bypass grafting.

Case Description

A 43-year-old man with chest pain during exertion for several months visited our institution. Physical examination revealed no signs of congestive heart failure. Electrocardiography was normal, but X-ray showed significant cardiomegaly. Transthoracic echocardiography showed normal left ventricular function with an ejection fraction of 60% and no findings of mitral regurgitation or asynergy. Contrast-enhanced computed tomography (CT) revealed that the left anterior descending artery (LAD) originated from the pulmonary artery (PA; ▶Fig. 1). The left circumflex artery (Cx) originated from the aortic root, similar to the normal left main coronary artery. The right coronary artery (RCA) origi-

nated from the aortic root as usual but was abnormally dilated. Angiography showed that the LAD was dilated, tortuous and filled by collaterals from the RCA (▶Fig. 2). In addition, the LAD drained to the PA. Myocardial scintigraphy showed redistribution of the anteroseptum wall, suggesting myocardial ischemia in the territory of the LAD. The patient was therefore diagnosed with ALADPA resulting in angina and underwent surgical repair with on-pump beating coronary artery bypass grafting (CABG).

The patient underwent a median sternotomy. The left internal thoracic artery (LITA) was carefully harvested. Cardiopulmonary bypass was established with aortic and right atrial cannulation. Similar to the preoperative diagnosis, the LAD was found to be dilated and originating from the anterior wall of the main pulmonary artery (▶Fig. 3A). The origin was easily peeled and double ligated with silk ties in two places (▶Fig. 3B). Thereafter, an arteriotomy was performed on the LAD, but there was heavy bleeding from the arteriotomy site due to collaterals from the RCA that became uncontrollable; therefore, the ascending aorta was clamped, and cardioplegic solution was administered. Under cardiac arrest, the LITA was anastomosed to the dilated LAD. The postoperative course was uneventful. The postoperative contrast-enhanced CT was performed on the seventh postoperative day and it showed that

received
July 7, 2020
accepted after revision
October 13, 2020

DOI <https://doi.org/10.1055/s-0040-1721470>.
ISSN 2194-7635.

© 2021. The Author(s).

This is an open access article published by Thieme under the terms of the Creative Commons Attribution-NonDerivative-NonCommercial-License, permitting copying and reproduction so long as the original work is given appropriate credit. Contents may not be used for commercial purposes, or adapted, remixed, transformed or built upon. (<https://creativecommons.org/licenses/by-nc-nd/4.0/>)

Georg Thieme Verlag KG, Rüdigerstraße 14, 70469 Stuttgart, Germany

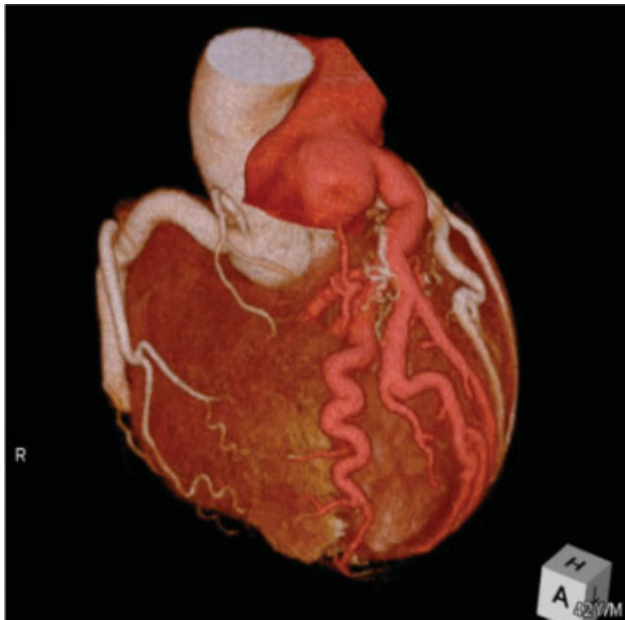


Fig. 1 Preoperative finding of coronary CT angiography. The left anterior descending artery was abnormally dilated and originated from the pulmonary artery. CT, computed tomography.

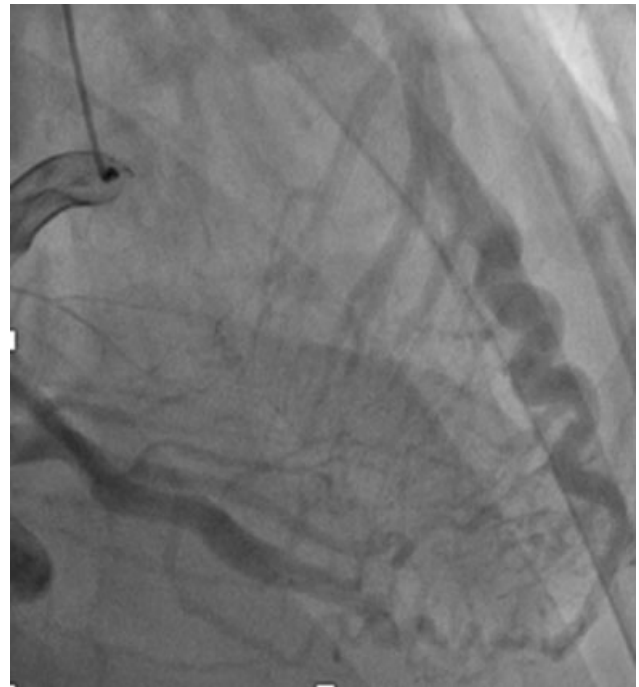


Fig. 2 Angiography of the right coronary artery. The left anterior descending artery was visualized through the collateral vessels from the right coronary artery.

the bypass graft was patent. After postoperative rehabilitation for 1 month, as usual in our hospital, the patient went home on the 34th postoperative day. Two years after the operation, the patient is doing well without any chest pain and follow-up CT-angiography revealed the graft was still patent (→ Fig. 4).

Discussion

In the present case, ALADPA was successfully treated with CABG using an LITA graft.

ALCAPA is a rare coronary anomaly, accounting for 0.24 to 0.4% of all congenital heart disease.² In some patients with ALCAPA, the LAD independently arises from the pulmonary artery, a variant called ALADPA, it is extremely rare to diagnose

ALADPA in an adult because patients with ALADPA usually become symptomatic in the first year after birth. Until now, there have been only a few case reports of ALADPA in adults.^{1,3}

In patients with ALADPA, the pressures in the coronary and pulmonary arteries are same in fetal life. However, after birth, the pulmonary artery pressure gradually decreases, causing ischemia of the LAD territory because of low LAD flow. Then, over time, collaterals from the other coronary arteries, normally from the RCA through the septal branches, result in LAD perfusion, depending on the collaterals. Zhang and colleagues stated that the development of collateral circulation from the RCA to the LAD determines the extent of myocardial ischemia,

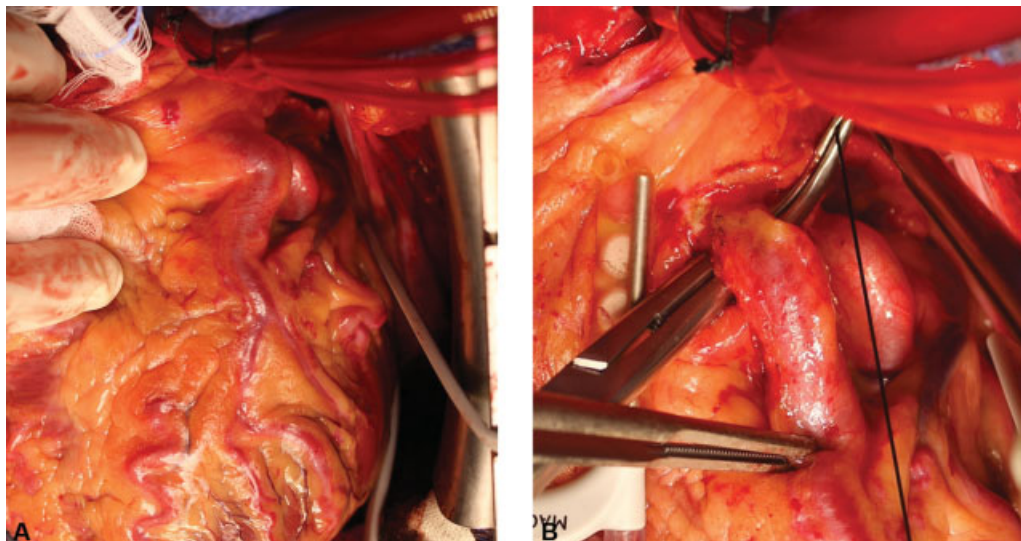


Fig. 3 Intraoperative findings: (A) the whole view of the left anterior descending artery (LAD). (B) The proximal part of LAD was ligated twice.

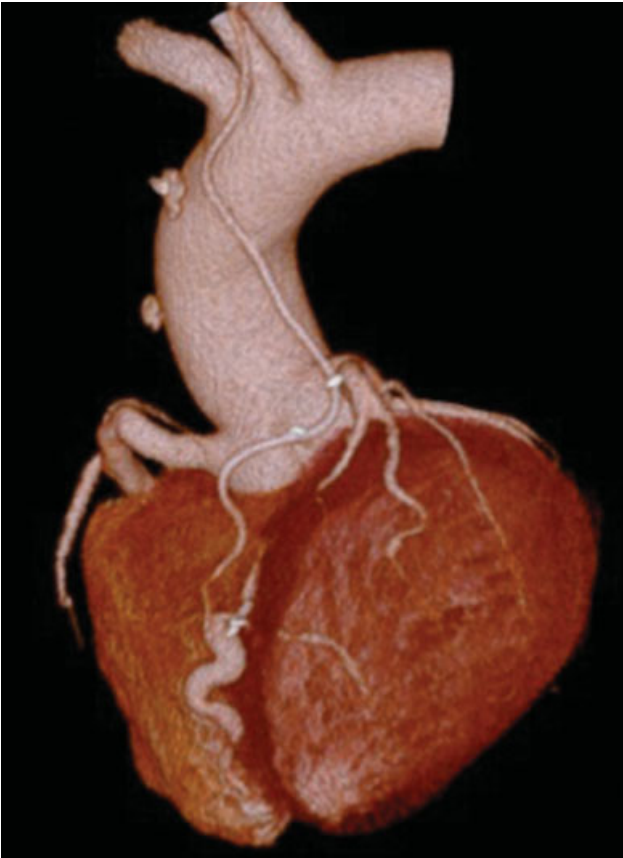


Fig. 4 Postoperative finding of coronary CT angiography. Two years after the operation, the graft was still patent. CT, computed tomography.

and the collaterals from the RCA to the LAD are well established in adult patients with ALADPA.³ Additionally, Rajbanshi and colleagues reported that collateral flow from the RCA is shunted to the pulmonary artery through the LAD that causes coronary steal phenomenon.⁴ Therefore, even in patients with ALADPA who survive to adulthood, chronic ischemia, heart failure, and sudden death can occur because of mitral regurgitation and ventricular arrhythmia. Thus, surgical repair should be performed in asymptomatic patients at the time of diagnosis.

The surgical strategy is to establish a two-coronary system, as in cases of ALCAPA. Rajbanshi and colleagues stated that simple ligation called a single coronary system increased mortality.⁴ There are several surgical strategies to establish the two-coronary system in adults which are the direct implantation of the coronary artery into the aorta and CABG. Jujjavarapu and colleagues reported that in direct implantation, it can be technically difficult to mobilize the LAD to reach the aorta in adults.⁵ In fact, direct implantation would have been anatomically difficult in this case because the LAD originated from the anterior wall of the main pulmonary artery. In addition, they also stated that total arterial revascularization with off-pump coronary artery bypass grafting and ligation of the origin of the left main stem in adults with ALCAPA is a safe and reasonable alternative procedure.⁵ Thus, CABG may be the choice of treatment in adult ALADPA patients in whom mobilization of the LAD is technically difficult. Tauchi and colleagues

suggested that the saphenous vein was more useful for the maintenance of flow than the internal thoracic artery (ITA).⁶ However, arterial grafting with the ITA can provide better long-term outcome than with a vein graft in general and it may be similar also in ALCAPA patients if short-term patency could be ensured against flow competition. Although ITA patency might be affected by competitive flow from collaterals, Kawasuji and colleagues stated that ITA was acceptable for a moderately stenotic coronary artery.⁷ In this case, the ITA graft was patent on postoperative CT, although the LAD was dilated and competitive flow was confirmed. Retrospectively, our decision to perform ITA grafting was not the false one. Additionally, Minamida and colleagues reported that a patient underwent redo CABG using ITA for postoperative vein graft stenosis with ALCAPA.⁸

Conclusion

ALADPA in adults is an extremely rare coronary anomaly. To avoid heart failure and sudden death, surgical repair should be performed in asymptomatic patients with ALADPA. Although the surgical strategies are controversial, we recommend establishing a two-coronary system with CABG. Although ITA patency might be affected by competitive flow from collaterals, our experience suggests that ALADPA could be adequately treated with CABG using ITA in selected patients.

Conflict of Interest

None declared.

References

- 1 Madhavan S, Sathish G, Jayaprasad N. ALADPA: Snake in the heart. *NHJ* 2015;12:27–30
- 2 Russo V, Musuraca AC, La Palombara C, Di Pasquale G, Fattori R. Cardiovascular images. Bland-White-Garland syndrome discovered in an elderly man. *Circ Cardiovasc Imaging* 2009;2(03):276
- 3 Zhang Y, Wang B, Li Y, Xie M. A rare case of anomalous origin of the left anterior descending artery from the pulmonary artery. *Echocardiography* 2019;36(06):1208–1210
- 4 Rajbanshi BG, Burkhart HM, Schaff HV, Daly RC, Phillips SD, Dearani JA. Surgical strategies for anomalous origin of coronary artery from pulmonary artery in adults. *J Thorac Cardiovasc Surg* 2014;148(01):220–224
- 5 Jujjavarapu KR, Salhiyyah K, Witzke H, Amrani M. Off-pump total arterial revascularization for anomalous origin of the left coronary artery from the pulmonary artery. *Ann Thorac Surg* 2013;96(06):2234–2236
- 6 Tauchi Y, Yasuda T, Yajima S et al. Surgical treatment in an adult ALCAPA case. *Sinzo*. 2013;45:579–584
- 7 Kawasuji M, Sakakibara N, Takemura H, Tedoriya T, Ushijima T, Watanabe Y. Is internal thoracic artery grafting suitable for a moderately stenotic coronary artery? *J Thorac Cardiovasc Surg* 1996;112(02):253–259
- 8 Minamida T, Sugiki T, Itou M, Kamikubo Y, Takahira M. [Redo off-pump coronary artery bypass for postoperative vein graft stenosis of anomalous origin of the left coronary artery from the pulmonary artery; report of a case]. *Kyobu Geka* 2017;70(10):859–862