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Case Report

Successful off-pump reimplantation of the anomalous right coronary artery from the pulmonary artery: A case report

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ABSTRACT

Anomalous origin of the right coronary artery from the pulmonary artery (ARCAPA) is a rare occurrence that requires surgical repair, typically via cardiopulmonary bypass (CPB). In this study, we present the case of a patient with ARCAPA with a high risk of cerebral infarction and left main trunk stenosis. However, because of the high risk of cerebral infarction, CPB was no longer an option during surgical intervention. Instead, we performed off-pump reimplantation of the ARCAPA to the ascending aorta and coronary artery bypass grafting of the left coronary artery. The patient had an uneventful postoperative course. Based on the successful outcomes of this case, we suggest off-pump reimplantation of the ARCAPA to the ascending aorta as a useful alternative for patients who are not eligible to undergo CPB during surgical repair.

<Learning objective: Although surgical repair of the anomalous origin of the right coronary artery from the pulmonary artery (ARCAPA) usually requires cardiopulmonary bypass (CPB), we present a successful off-pump reimplantation for those who are not eligible to undergo CPB. We performed off-pump reimplantation of the ARCAPA to the ascending aorta and coronary artery bypass grafting of the left coronary artery. Therefore, we found off-pump reimplantation of the ARCAPA to the ascending aorta to be a useful alternative to CPB in high-risk patients.>

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Introduction

Anomalous origin of the right coronary artery from the pulmonary artery (ARCAPA) is rare; its incidence is 0.002% and it is estimated to comprise 0.12% of coronary anomalies [1]. ARCAPA can be diagnosed during adulthood, especially if the blood supply from the left coronary artery (LCA) is sufficient for the whole heart and the shunt flow exists through the ARCAPA. In most cases, surgical repair of the ARCAPA is achieved through cardiopulmonary bypass (CPB); however, the patient in this report had a high risk of cerebral infarction as the left internal carotid artery (LICA) and right middle cerebral artery (RMCA) had chronic total occlusion. Thus, the use of CPB was

considered high risk and as an alternative, we performed off-pump re-implantation of the ARCAPA to the ascending aorta in addition to off-pump coronary artery bypass grafting (CABG) for the LCA lesion.

Case report

A 53-year-old woman complained of shortness of breath during exertion for the past 3 months in the year 2008. A provisional diagnosis of New York Heart Association (NYHA) class II degree heart failure was made, and coronary angiography was performed to investigate the cause. Consequently, the patient was diagnosed with ARCAPA. However, ischemia was not observed on myocardial scintigraphy. The treatment plan was to administer oral medications and observe the clinical course of the patient. Subsequently, in 2018, the patient experienced chest pain and dyspnea during exertion. A decision was made to conduct a causal test based on the diagnosis of NYHA class II heart failure. In addition, the patient had

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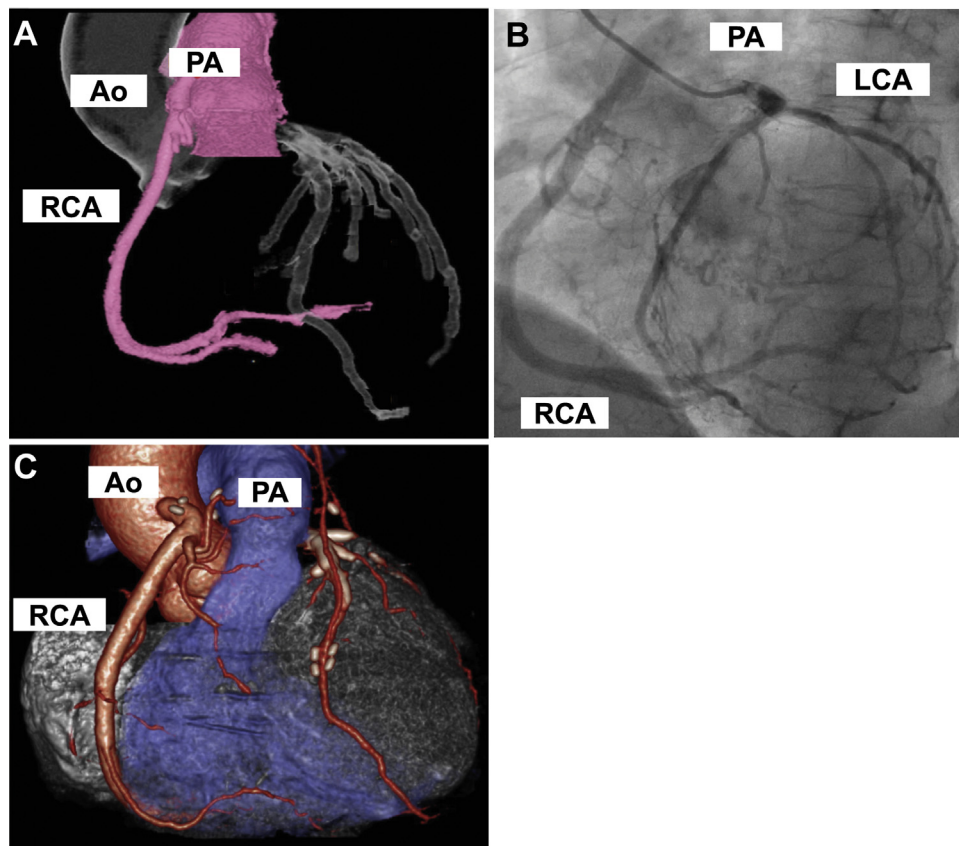


Fig. 1.

Preoperative coronary computed tomographic reconstruction shows the ARCAPA arising from the main pulmonary artery (A). (B) Preoperative coronary angiography. (C) Postoperative coronary computed tomographic reconstruction.
Ao, ascending aorta; ARCAPA, anomalous origin of the right coronary artery from the pulmonary artery; PA, pulmonary artery; RCA, right coronary artery; LCA, left coronary artery.

a past medical history of hypertension. Informed consent was obtained from the patient before the surgery for the publication of this report.

Preoperative echocardiographic examination showed normal left ventricular dimensions (end-diastolic diameter, 45 mm; end-systolic diameter, 30 mm), preserved function (ejection fraction, 63%), and mild mitral regurgitation. Myocardial scintigraphy revealed inferolateral wall ischemia and coronary angiography showed 90% stenosis of the left main trunk (LMT) with calcification. Although the ARCAPA showed no significant stenosis, it drained blood into the main pulmonary artery (MPA), which was visualized with retrograde flow through collaterals from the left anterior descending artery (LAD).

Based on the coronary computed tomography angiography (CTA), we judged that reimplantation of the ARCAPA was possible, with sufficient length to connect to the ascending aorta (Fig. 1). Thus, we planned to perform an off-pump reimplantation of the ARCAPA to the ascending aorta and off-pump CABG for LCA. Surgery was performed through median sternotomy under general anesthesia.

We dissected the origin of the ARCAPA from the MPA (Fig. 2) and the proximal region of the ascending aorta. We ensured that reimplantation of the ARCAPA to the ascending aorta was anatomically possible without CPB, provided that the total length of the ARCAPA was used. We first performed the off-pump CABG, connecting the left internal mammary artery (IMA) to the LAD and the right IMA to the obtuse marginal branch.

Preoperative coronary angiography showed that collateral circulation from the LAD into RCA was significant (Fig. 2). This could cause ischemia in both LAD and RCA territories during the anastomosis between the LAD and LITA. Therefore, we carefully inserted an intra-coronary shunt tube into the LAD to maintain collateral flow into the RCA. Consequently, there was no change in the vital signs during the anastomosis between the LITA and LAD, and the patient's condition was stable.

We placed a purse string suture around the origin of the ARCAPA to salvage the maximum length of the ARCAPA. Thereafter, we transected the ARCAPA at 2 mm distal to its origin from the MPA. The hole in the MPA was temporarily closed using the purse-string suture. It was then sutured with the horizontal mattress and over-and-over sutures, using the 5-0 monofilament suture. Finally, the ARCAPA was reimplanted to the ascending aorta at the level of the sinotubular junction in a side-to-end anastomosis using Enclose II® (Novare Surgical Systems, Inc., Cupertino, CA, USA) (Fig. 2).

Coronary CTA on postoperative day 5 showed that both the IMA and reimplanted ARCAPA were patent (Fig. 1). The patient was discharged on postoperative day 14, with a favorable 9-month outpatient follow-up.

Discussion

We report the case of a patient with ARCAPA in whom off-pump reimplantation of the ARCAPA to the ascending aorta and

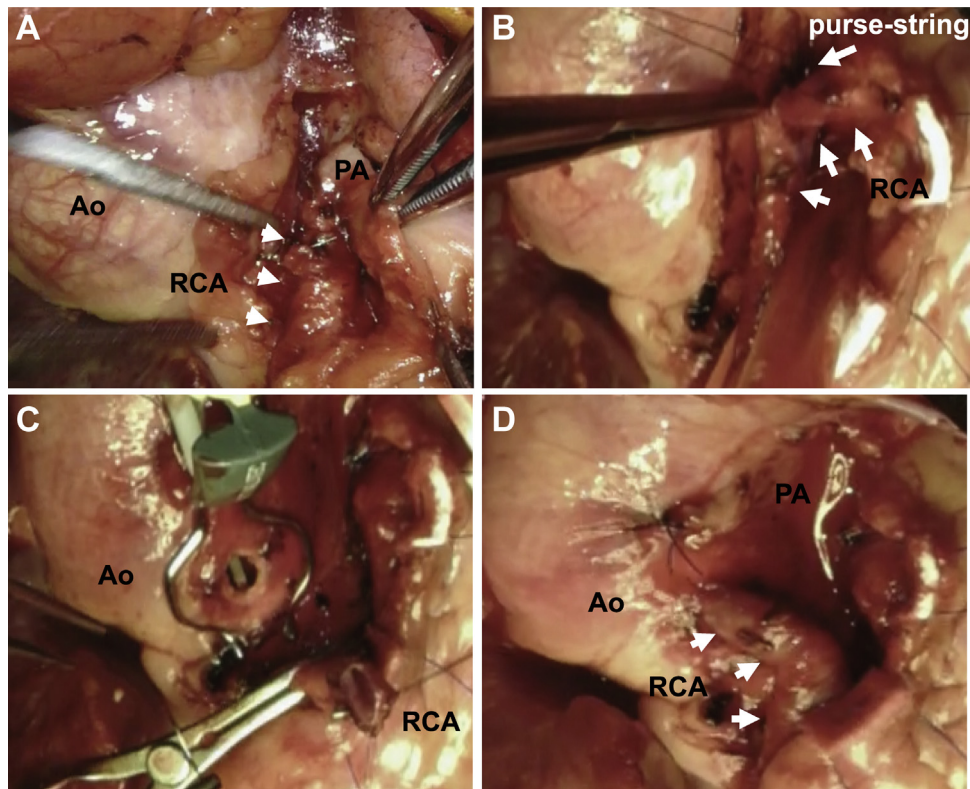


Fig. 2.

Operative photograph showing reimplantation of the ARCAPA into the aorta. White arrows show the ARCAPA. (A) The ARCAPA arising from the main pulmonary artery. (B) The purse-string for ARCAPA. (C) Anastomosis of ARCAPA to Ascending Aorta using Enclose II®. (D) The reimplanted ARCAPA into the aorta. Ao, ascending aorta; ARCAPA, anomalous origin of the right coronary artery from the pulmonary artery; PA, pulmonary artery; RCA, right coronary artery.

off-pump CABG for the LCA were performed, with positive results. The incidence of ARCAPA is reported to be 0.002% in the general population; approximately 0.12% of all coronary anomalies [1]. Usually, these patients remain asymptomatic and survive into adulthood, although sudden death caused by a cardiac event has been reported [1]. A murmur is the most frequent symptom while the clinical presentation of patients with ARCAPA is usually atypical and includes dyspnea (17%), fatigue (13%), congestive heart failure (30%), angina (17%), myocardial infarction (9%), and even sudden cardiac arrest (17%) [1].

The coronary flow reserve of an ARCAPA depends on the collateral flow from the LCA system; therefore, significant stenosis in the LCA has a greater impact, causing ischemia in the right coronary artery (RCA) region as well as the LCA region.

There are three surgical options for ARCAPA: (1) reimplantation of the ARCAPA to the ascending aorta; (2) ligation of the ARCAPA with CABG for an ARCAPA with a saphenous vein graft (SVG); and (3) simple ligation of the ARCAPA [2]. Reimplantation of the ARCAPA to the ascending aorta is the ideal option because it leads to establishment of double coronary circulation. This surgical repair allows for normalization of the coronary flow reserve. CABG for an ARCAPA with an SVG should be selected when reimplantation is not possible such as in a situation wherein the ARCAPA originates from the posterior wall of the pulmonary artery. The drawback of CABG is the long-term patency of the SVG in young patients. Simple ligation of the ARCAPA should not be selected unless both reimplantation and CABG are not possible. Coronary

CTA provides details regarding the location and course of an ARCAPA. It is helpful for the surgeon to decide whether reimplantation is possible.

To the best of our knowledge, there are 8 reports of 9 cases [3–10] (Table 1) in the literature after 2010. The mean age of the patients was 43.6 years, and 6 men were reported to have suffered from this condition. Of the 9 cases, 6 patients (66.6%) underwent reimplantation of the ARCAPA to the aorta and 3 (33.4%) underwent ligation of the ARCAPA with CABG. Within the 3 cases, 1 used the SVG, 2 used the right IMA. All cases showed a favorable postoperative course. No case of simple ligation of the ARCAPA has been reported since 2010.

Our patient presented with chronic total occlusion of the LICA and the RMCA; due to these lesions, the use of CPB was considered unsuitable. Therefore, we performed an off-pump surgery. The challenge of off-pump reimplantation is the need to transect the ARCAPA close to its origin in order to salvage its entire length. Therefore, we placed purse-string sutures around the origin of the ARCAPA to transect it at the most proximal site so the utilization of the entire length of the ARCAPA was possible. In addition, the Enclose II® anastomosis device was useful for proximal anastomosis of the ARCAPA when we performed the off-pump technique. Applying the side-clamp of the ascending aorta at the proximal site was difficult in the off-pump procedure due to the bulging right ventricular outflow.

In conclusion, based on our findings and experience, the off-pump reimplantation of the ARCAPA to the ascending aorta is useful for high-risk patients who cannot undergo CPB.

Table 1 Review of 8 reports that show surgeries of 9 patients with anomalous origin of the right coronary artery from the pulmonary artery.

Case	Year	Reference	Age	Gender	Operation	Use of CPB	Hospital death	Complication	Follow up period
1	2011	Parasramka et al. [3]	21	M	Reimplantation of the RCA	n/d	-	None	n/d
2	2013	Kuba et al. [4]	71	M	CABG (LITA-LAD) + reimplantation of the RCA	+	-	None	11 years
3	2014	Rajbanshi et al. [5]	78	F	Reimplantation of the RCA	+	-	None	10 years
4	2014	Rajbanshi et al. [5]	34	M	Reimplantation of the RCA	+	-	None	0.8 years
5	2015	Chernogrivov et al. [6]	17	M	Reimplantation of the RCA	+	-	None	3 months
6	2018	Rawala et al. [7]	57	M	Reimplantation of the RCA	n/d	-	None	n/d
7	2014	Izgi et al. [8]	21	M	CABG (RIMA-RCA) + RCA ligation	n/d	-	None	n/d
8	2015	Fuglsang et al. [9]	39	F	CABG (RIMA-RCA) + RCA ligation	n/d	-	None	n/d
9	2017	Balakrishna et al. [10]	55	F	CABG (SVG-RCA) + RCA ligation	n/d	-	None	n/d

In cases 2, 3, 4, and 5, the RCA was reimplanted as the button with some pulmonary artery wall. In cases 1 and 6, the manner of reimplantation of the RCA was not described. RCA, right cardiac artery; CABG, coronary artery bypass grafting; CPB, cardiopulmonary bypass; LITA, left internal mammary artery; LAD, left anterior descending artery; SVG, saphenous vein graft; RIMA, right internal mammary artery; n/d, not described.

Conflict of interest

The authors declare that there is no conflict of interest.

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