

Does Coronary Artery Bypass Surgery still have a Role in the Management of Patients with Anomalous Origin of the Right Coronary Artery from the Left Sinus of Valsalva?

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Abstract

Objective: An anomalous origin of the Right Coronary Artery (RCA) from the left sinus of Valsalva with an inter-arterial course is a rare congenital anomaly. We aimed to assess mid-term results after off-pump coronary artery bypass grafting for treatment of Anomalous right coronary artery arising from the left sinus of Valsalva.

Methods: Between the year 2008 and 2012, a total of 16 patients underwent off-pump CABG for treatment of the right coronary artery of an anomalous origin from the Left Sinus of Valsalva. Their details such as risk factors, operative details, and results of multi-slice coronary angiography follow-up were assessed.

Results: The mean age of these patients was 34.8 ± 4.68 years, and most of the patients were male (15/16: 93.75%). The patients received a mean of 1.5 ± 0.87 grafts. Closure of the RCA was completed for all patients except one who developed ischaemic symptoms upon closure of the proximal part of the RCA (15/16). We had one mortality. In the remaining patients, Symptoms persisted in 2/15 patients. The mean follow-up time was 63.4 ± 28.6 months. All patients had patent vessels on five years follow-up Multi-slice computed tomography scan except for one patient who showed RCA graft occlusion on 1-year follow-up scan.

Conclusion: Off-pump CABG of RCA with anomalous origin can be done safely with excellent early and midterm results. Proximal RCA ligation is an essential step to the success of the CABG, but intraoperative challenge testing is required to confirm sufficiency of the ITA graft to re-perfuse the supplied territory.

Keywords: Coronary artery anomaly; Inter-arterial course; Coronary CT, Bypass grafting

Abbreviations CABG: Coronary Artery Bypass Grafting; CPB: Cardiopulmonary Bypass; CT: Computed Tomography; ITA: Internal Thoracic Artery; RCA: Right Coronary Artery; RITA: Right Internal Thoracic Artery; SVG: Saphenous Vein Graft; MSCT: Multi Slice Computed Tomography

Introduction

An anomalous origin of the Right Coronary Artery (RCA) from the left sinus of Valsalva with an inter-arterial course is a rare congenital anomaly reported in 0.05% to 0.1% of patients undergoing coronary artery angiography. The most severe presentations of this anomaly are sudden acute myocardial infarction and sudden cardiac death; these presentations are most common in young healthy athletes. The other clinical presentations are highly variable, ranging from asymptomatic to variable symptomatology, including dyspnoea, arrhythmia, myocardial ischemia, and exertional syncope. Previous studies have assumed that the various clinical manifestations are related to variable individual degrees of external exertional systolic luminal compression of the RCA (Right Coronary Artery) by the great vessels due to its inter-arterial course [1-3].

Previous studies have reported that coronary artery anomalies are the second most frequent cause of sudden death in young athletes. Therefore, active management of these patients is recommended to improve their quality of life and to prevent sudden death.

In symptomatic patients, medical treatment is supplemented with surgical treatment. Many surgical techniques, including re-implantation of the ostium to the correct sinus from outside the aorta, un-roofing of the RCA from inside the aorta, and Coronary Artery Bypass Grafting (CABG) using an internal thoracic artery or saphenous vein graft, have been developed to correct these anomalies [4].

In this series, we present our surgical experience with off-pump CABG and ligation of the proximal RCA. Early and mid-term follow-up results for a mini-series of 16 patients with an anomalous origin of the RCA from the left sinus of Valsalva and an inter-arterial course (coursing between the aorta and pulmonary artery) were recorded.

Materials and Methods

A retrospective study was conducted on 16 patients with an anomalous RCA arising from the left sinus of Valsalva who underwent surgical correction between Jan 2008 and Dec 2012 in Ain Shams University and affiliated hospitals.

Data collection

After the approval of the ethical committee for research activities in Ain Shams University, a review of the cardiac surgical database was done and the following data were collected. Preoperative data included age, sex, weight, symptomatology, and risk factors such as diabetes mellitus, hypertension, and dyslipidemia. Preoperative investigations included coronary angiography or Multi-Slice Computed Tomography (MSCT) angiography with the determination of arterial dominance and the number of diseased vessels. Dobutamine stress echocardiography and stress thallium scintigraphy were also included to describe the ischemic patterns in relation to exercise and stress. Surgical details, the use of internal thoracic arteries, the number of grafts, and ligation of the proximal RCA segment were recorded. We also recorded the patency of the grafts according to MSCT angiography during the follow-up period. Post-operative challenge tests were conducted by stress dobutamine echocardiography for recurrent signs of ischemia.

Operative technique

Under normothermic conditions, general anesthesia and a muscle relaxant were administered, followed by traditional median sternotomy with harvesting of the right and/or left internal thoracic artery and the venous conduits. Heparin was given at a dose of 1-1.5 mg/kg, which is one-third of the dose used for open heart procedures, to achieve an Activated Clotting Time (ACT) of 300 seconds, which was regularly assessed every 30 minutes. Anastomosis was carried out in the sequence of the left anterior descending artery, its diagonal branch, the right coronary artery and the obtuse marginal artery with every distal anastomosis followed by the corresponding proximal anastomosis. Performing the proximal anastomosis after each distal anastomosis ensures immediate perfusion of the selected vessel, thus lessening the ischaemic time and decreasing myocardial injury. The order of revascularization was selected based on the principle that increased cardiac displacement can be tolerated when more regions are revascularized, as greater displacement is required for the lateral wall followed by the inferior walls. During the RCA anastomosis, the table was levelled or placed in the Trendelenburg position with the left pericardial retraction sutures relaxed to allow the heart to tilt and fall into the left thoracic cavity. Temporary pacing was used as necessary, as transient occlusion of the RCA can cause ischemia in the AV node. Stabilization was achieved using the Medtronic octopus stabilizer

(Medtronic Inc., Minneapolis, USA). The more distal part of the RCA was the preferred site of graft anastomosis. After establishing the anastomosis to the RCA, snaring of the RCA proximal segments using 3/0 Prolene suture is done. Electrocardiography (ECG) was recorded, and cardiac functions were assessed. Low dose dobutamine was initiated to increase cardiac stress. Cardiac functions were assessed by continuous trans-esophageal echo and ECG monitoring. In the absence of changes in cardiac function or induced ischemia, the snare was completed for total ligation of the artery, and the procedure was considered complete. Closure of the proximal RCA was completed using Prolene 2/0 sutures to avoid competitive flow and graft occlusion.

Follow-up

Follow-up was conducted by following patient symptomatology for recurrence. MSCT coronary angiography was performed as a screening test to determine the patency of the grafts. CT was done routinely at five years following CABG and on indications suggestive of recurrent ischemia. Repeat stress testing was also performed to assess symptom recurrence *via* stress dobutamine echo.

Statistical data analysis

The patient data were coded as variables. Quantitative variables are expressed as the mean and SD. Qualitative variables are expressed as frequencies and percentages. The collected data were reviewed using the Statistical Package for Social Science (IBM SPSS Statistics for Windows, Version 20.0. Armonk, NY: IBM Corp, 2011).

Results

From Jan 2008 to Dec 2012, approximately 3,200 CABG procedures were performed at Ain Shams University hospitals in Cairo, Egypt. During this period, a total of 16 patients with an anomalous origin of the RCA from the left sinus of the aortic root with an abnormal inter arterial course were treated (Table 1). The mean age of these patients was 34.81 ± 4.68 years, and most of the patients were male (15/16: 93.75%). The mean weight was 90.875 ± 8.9 kg. A total of 8/16 (50%) of the patients were hypertensive, 6/16 (37.5%) were diabetic, 3/16 (18.75%) were hyperlipidaemic, 11/16 (68.75%) presented with atypical chest pain symptomatology, 6/16 (37.5%) presented with exertional syncope, and 8/16 (50%) presented with palpitations.

S. No.	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16
Year	2008	2008	2009	2010	2010	2010	2011	2011	2011	2011	2012	2012	2012	2012	2012	2012
Age (years)	35	30	40	32	31	30	30	36	45	40	35	32	33	31	35	42
Weight (kilograms)	90	80	103	85	89	100	80	95	80	85	85	100	87	90	110	95
Sex	M	M	M	M	M	M	M	M	M	M	M	M	F	M	M	M
Hypertension	-	-	+	-	-	+	-	+	+	-	-	+	+	+	+	+
Diabetes	-	-	+	+	-	-	+	-	+	-	-	+	-	-	+	-
Dyslipidaemia	+	-	+	-	-	-	+	-	-	-	-	+	-	-	-	-
Chest pain																

(exercise-induced) Typical angina	-	-	+	-	-	-	+	-	+	-	-	+	-	-	+	-
Atypical angina	+	+	-	+	+	+	-	+	-	+	+	-	+	+	-	+
Syncope	+	-	-	+	-	-	-	+	-	-	-	+	-	-	+	+
Palpitations	+	+	+	-	-	+	-	+	-	+	+	-	-	+	-	-
dobutamine stress echography	+	+				+		+		+			+			+
Preoperative Stress thallium scintillography	-	-	-	+	+	-	-	-	-	-	+	-	-	+	-	-
Surgery																
Number of diseased Vessels	1	1	2	1	1	1	3	1	2	1	1	3	1	1	2	1
Right Coronary artery dominance	+	+	+	-	-	+	+	-	-	+	+	+	+	+	+	+
Number of grafts	1	1	2	1	1	1	4	1	2	1	1	3	1	1	2	1
RITA to RCA grafting	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Closure of proximal RCA	-	+	+	+	+	+	-	+	+	+	+	+	+	+	+	+
Post-surgery med.	+	-	+	-	-	-	-	-	+	-	-	+	-	-	+	-
Post-procedure recurrent symptomatology	+	-	+	-	-	-	-	-	-	-	-	-	-	-	-	-
Mortality	-	-	-	-	-	-	+	-	-	-	-	-	-	-	-	-
Duration of follow-up (months)	12	100	30	108	80	70	0	73	75	72	70	85	60	61	59	60
CT follow-up of RCA graft patency																
CT at 1 year	O	P	p	p	P	P	-	P	P	P	P	P	P	P	P	P
CT at 5 years	-	P	-	P	P	P	-	P	P	P	P	P	P	P	P	P
Stress echocardiography at 6 months	+	-	+	-	-	-	-	-	-	-	-	-	-	-	-	-

Table 1: Patient characteristics and details.

The preoperative stress dobutamine echo was positive in 7/16 patients (43.75%). Preoperative Stress thallium scanning was positive in 4/16 (25%) patients. The mean number of diseased vessels on coronary angiography was (1.4 ± 0.7). Coronary circulation was right-dominant in 75% (12/16) of the patients. Right Internal Thoracic Artery (RITA) grafting of the RCA was used in all patients. The patients received a mean of 1.5 ± 0.87 grafts.

Closure of the RCA was completed for all patients except one (patient No.1) who developed ischaemic symptoms upon closure of the proximal part of the RCA (15/16).

Symptoms persisted in 2/16 patients (patients no. 1 and 3). The first patient exhibited RCA occlusion on one year. Follow up MSCT angiography, and the second patient had diabetes, dyslipidemia, and extensive two vessels disease.

All remaining patients were asymptomatic following the procedure, with negative stress test results on dobutamine echo done six months following surgery.

We had one mortality (patient 7) in the series. The patient was a 30 years old male with diabetes and dyslipidemia. The patient had extensive three vessels disease with atypical RCA origin from the left aortic valve sinus. The patient presented with typical exercise-induced anginal pain. The Patient underwent off-pump CABG with LITA to LAD, SVG (Saphenous Vein Graft) to OM (Obtuse Marginal) artery, RITA to RCA without ligation of the proximal part of the RCA. Following surgery, the patient suffered from low cardiac output and died on the 4th postoperative day.

The mean follow-up time was 63.4 ± 28.6 months. All patients had patent vessels on five years. Follow up MSCT (Figure 1), except for one

patient (patient No.1) who showed RCA graft occlusion on one-year follow-up examination. The patient was a 35 years old male with dyslipidemia. He presented with atypical angina, syncope, and palpitations. He underwent off-pump single RITA to RCA grafting without ligation of the origin of the RCA. Symptoms persisted after the procedure. MSCT angiography was done one year following surgery and demonstrated an occluded graft. The patient underwent repeat on-pump CABG with endarterectomy of the RCA and LITA (Left Internal Thoracic Artery) to a saphenous venous overlay patch to the endarterectomized vessel. The patient died after the procedure from severe right ventricular dysfunction on the 2nd postoperative day.

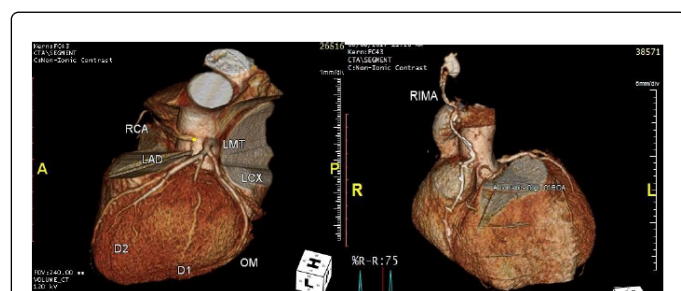


Figure 1: Multi-slice CT of patient two before surgery-to the left-and after surgery-to the right-with the RITA graft.

Discussion

Coronary artery anomalies constitute a heterogeneous group of malformations. Anomalies of a coronary artery may involve the origin, course, or terminus of the artery, and these anomalies present an incidence of 0.61% to 5.64% on coronary angiography. One particular category of these anomalies involves an abnormal origin of the RCA from the left aortic sinus (Figure 2), which has an incidence of 0.12% to 0.92%. This specific category of coronary artery anomaly involves an inter-arterial course of the RCA between the great vessels before it emerges on the heart surface.

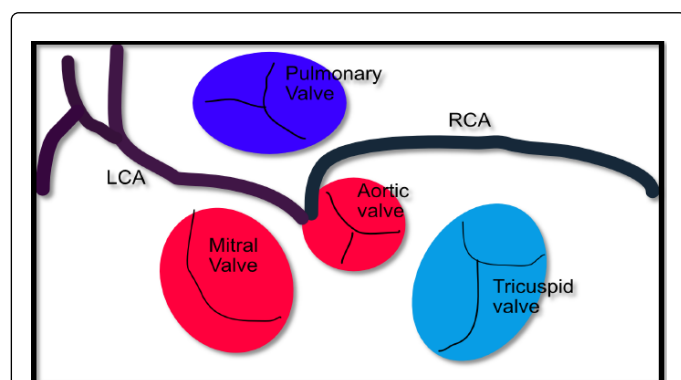


Figure 2: Diagram showing the anatomy of the right coronary artery originating from the left aortic sinus with an inter-arterial course.

Patients with this form of anomaly are vulnerable to ischemia and sudden death. Three mechanisms have been proposed to explain this increased susceptibility to ischaemic events (Figure 3). The first mechanism is the sharp angulation and kinking of the RCA as it emerges from the opposite sinus. The coronary ostium morphology,

which changes to a slit-like opening, is more likely to develop a valve mechanism. External compression of the abnormal segment of the artery between the great vessels can occur, especially during strenuous exercise, as the increased flow and expansion of the great arteries induced by exercise add to the insult. This occurs more often in young patients, as they have healthy and distensible vessels. If the RCA is the dominant vessel, then ischaemic events are augmented and can be fatal [1-4].

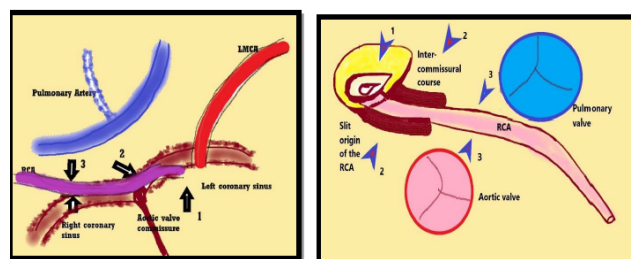


Figure 3: Diagram showing the mechanism of ischemia in the right coronary artery arising from the left aortic sinus with an inter-arterial course. Arrow 1 demonstrates the slit-like origin of the RCA in this case with the acute take-off angle. Arrow 2 shows the second mechanism with the intramural course near the aortic valve commissure. Arrow 3 demonstrates the external compression mechanism between the aorta and the pulmonary artery especially during systole and during systolic arterial expansion.

Based on previous pathophysiological processes, the clinical implications of coronary artery anomalies can be variable. Most patients are asymptomatic. However, atypical chest discomfort may be a prevalent symptom, prompting patients to seek medical attention. Some patients are identified by a positive stress test or signs of ischaemic heart disease on ECG [5-7]. Sudden death can be the first presentation, especially in young athletic patients with right-dominant coronary circulation [2,8,9].

Several treatment options are available for the right ACAOS with symptoms of ischemia, including conservative follow-up with frequent assessments and medical therapy. Interventional and surgical options are also available if these approaches fail [10].

According to the ACC/AHA guidelines published in 2008, patients can be defined as symptomatic and asymptomatic according to ischaemic symptoms. In asymptomatic patients, the primary objective is to improve the quality of life and to prevent sudden death. These individuals are advised against the heavy physical activity, strenuous exercise, and competitive sports. Medication with beta blockers is also proposed as a prophylactic measure [9,10]. The study of Maron et al. [9] reported zero sudden deaths in a 5-year follow-up of patients on beta blockers and without symptoms of ischemia.

The ACC/AHA guidelines strongly recommend surgery in patients with ischaemic symptoms based on level b evidence. Several surgical techniques are available for the treatment of right ACAOS. These techniques are un-roofing the origin of the right coronary artery vessel, re-implantation to the correct sinus, and Coronary Artery Bypass Grafting (CABG) [10,11].

The un-roofing procedure involves manipulation of the orifice and enlargement or reduction of the take-off angle of the coronary artery origin, which reduces lateral compression on the intramural part of the

coronary artery. This technique may require takedown and reattachment of the inter-coronary commissure, which could consequently affect aortic valve stability and cause post-procedure aortic insufficiency [12,13]. The long-term patency of the neo-ostium has not been sufficiently established. Moreover, this technique does not address the inter-arterial course of the coronary artery. This method has been adopted by many surgeons (Mustafa et al.) [3], but the long-term outcomes have not yet been confirmed.

Coronary excision and re-implantation have also been proposed. However, neo-ostium stenosis can occur and may result in the need for further surgical correction, including CABG [14].

CABG is technically straightforward and can be performed off-pump with easy access to the target anastomotic site. This approach eliminates the inherent risks of un-roofing or re-implantation, as there is no need to open the aorta or manipulate the aortic valve. However, this technique has two drawbacks. First, the patients are usually young, and there is a potential risk of requiring re-intervention due to a decline in long-term graft patency with aging. Second, the flow from the natural RCA ostium could become competitive, as the flow is unobstructed at rest, which could decrease the patency of the graft and cause graft atrophy [15-18]. This has led to a trend towards ligation of the artery proximal to the graft site [15]. The RITA (Right Internal Thoracic Artery) is an easy and valuable option for grafting the RCA in these patients, providing an arterial graft with the best long-term patency outcomes. Some authors have expressed doubt regarding the sufficiency of the RCA graft to provide adequate flow to cover the whole RCA territory due to the presence of leaks and advise intraoperative stress testing before occlusion of the proximal origin of the RCA to ensure graft adequacy. CABG is considered superior to stent implantation in this pathology [19,20]. As Percutaneous Coronary Intervention (PCI) with stent implantation relieves systolic compression, but PCI in abnormal vessels is challenging to perform. Furthermore, selective cannulation of the ostium of the RCA is technically demanding, as the RCA has a small ectopic slanting orifice. This procedure may be facilitated with an MSCT pre-cath evaluation and MSCT-guided cannulation [21].

Several studies have previously described CABG for this type of anomaly. The work of Izhar et al. [20] described Minimally Invasive Direct Coronary Artery Bypass (MIDCAB) using the RITA to the RCA with good postoperative results [22]. Shah et al. [22] described RITA grafting to the RCA with ligation of the proximal RCA [15]. Matsueda et al. [23] described on-pump RITA grafting to the RCA with ligation of the RCA and reported good postoperative results. Izgi et al. [24] described RITA grafting to the RCA with ligation of the RCA and reported good early results.

Our series demonstrates that CABG is a valuable option for these patients. Off-pump CABG was used in all patients of our series. The right internal thoracic artery was used in all patients for grafting of the RCA and yielded good early and mid-term results as assessed by MSCT angiography of graft patency (14/15 patients, 93.3% patency rate).

Ligation of the proximal part of the RCA can be readily performed, and we think that this is an important step to ensure less competitive flow and better graft patency in the follow-up period.

Most of the patients were RCA-dominant, which probably caused the significant ischaemic events that prompted the patients to seek treatment.

Ligating the origin of the RCA can determine the success of using CABG for the treatment of patients with a right coronary origin from the left sinus. Fedoruk et al. [25] presented a series of five patients and demonstrated that the patients who underwent CABG without ligation of the proximal RCA exhibited early occlusion of the graft and required further corrective surgery, primarily for un-roofing. The un-roofing technique is considered an anatomical repair of the anomalous coronary artery because it addresses stenosis of the RCA origin and relieves the primary pathophysiologic mechanism of abnormal flow in the anomalous RCA. In 2003, Sabik et al. [16] demonstrated that competitive flow from the proximal target vessel contributed significantly to graft failure and revealed a linear relationship between graft failure and the degree of competitive flow, which adds more evidence to support ligation of the proximal part of the RCA during CABG for an abnormal RCA.

Some authors expressed concern about proximal RCA ligation during CABG for the anomalous RCA as the ITA graft blood flow might not be enough to re-perfuse all the RCA territory thus increasing the incidence of the post CABG hypo perfusion ischemic syndrome. This might lead to recurrent symptoms, depressed right ventricular function and even mortality. In 2004, Berger et al. [17] published a study on the long-term patency of internal mammary artery grafts and the degree of stenosis of the native vessel. This group found an association between the absence of significant stenosis of the native artery and a high occlusion rate of the arterial bypass conduit. Their study revealed a negative association between the uses of the internal mammary artery in the grafting of native vessels with only mild or moderate stenosis [15-18].

In our series, we used the ligation technique described in the study of Shah et al. [22]. In this technique, the cardiac functions are challenged by dobutamine infusion during temporary proximal RCA ligation to test for changes in cardiac functions and resultant ischemia. The snare is completed to total ligation of the RCA only after negative stress testing during the RCA snaring.

Limitation of the study

The small number of patients precludes a definitive conclusion that CABG is better than other modalities for the treatment of coronary anomalies. A prospective randomized trial is necessary to achieve the statistical significance required for such a conclusion. Such trial is actually not feasible in rare congenital anomalies due to small sample size. Intraoperative coronary flowmetry is a recently introduced method for evaluating coronary flow by the new grafts. Unfortunately, this method was not available at the time of the study. It would have given a better index of coronary perfusion and flow dynamics.

Conclusion

Off-pump CABG of RCA with anomalous origin can be done safely with good early and midterm results. Proximal RCA ligation is an essential step to the success of the CABG, but intraoperative challenge testing is required to confirm sufficiency of the ITA graft to reperfuse the supplied territory.

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