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From Compression to Correction, Righting the Wrong Route: Successful Surgical Correction of a Malignant Anomalous Right Coronary Artery [RCA]

Amyrul Azman[1*], Yong KC[1], Norliza Othman[2], Vendargon SJ[1], Er CY[1], S Karupiah[1]

[1]Department of Cardiothoracic Surgery, Hospital Sultanah Aminah, Johor Bahru, Malaysia
[2]Department of Cardiac Radiology, Hospital Sultanah Aminah, Johor Bahru, Malaysia

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***Corresponding author:** Amyrul Azman
Department of Cardiothoracic Surgery, Hospital Sultanah Aminah, Johor Bahru, Malaysia

Abstract **Case Report**

Background: Anomalous right coronary artery [RCA] arising from the left coronary cusp [LCC] with an interarterial course is a rare but clinically significant congenital coronary anomaly. This anatomical variation, often termed a "malignant" RCA, is associated with an increased risk of myocardial ischemia, arrhythmias, and sudden cardiac death, primarily due to dynamic compression between the aorta and pulmonary artery. Surgical intervention is generally recommended for symptomatic patients or those with high-risk anatomical features, even in the absence of significant coronary artery stenosis. ***Case Presentation:*** We present the case of a 38-year-old male with underlying diabetes mellitus, hypertension, and dyslipidemia who experienced recurrent episodes of angina, palpitations, and syncopal attacks. His past medical history included a motor vehicle accident [MVA] in 2014, resulting in multiple fractures managed with internal fixation. Initial cardiovascular evaluation revealed Canadian Cardiovascular Society [CCS] class I angina and New York Heart Association [NYHA] class II symptoms. Electrocardiography [ECG] showed sinus rhythm, while stress myocardial perfusion imaging demonstrated a mild perfusion defect in the inferior wall, correlating with the RCA territory. Coronary computed tomography angiography [CTCA] revealed an anomalous RCA originating from the LCC with an interarterial course between the aorta and pulmonary artery. Despite the absence of significant obstructive coronary artery disease, the high-risk anatomy of the RCA and the patient's recurrent symptoms warranted surgical intervention. Following a multidisciplinary discussion, the patient underwent surgical translocation of the anomalous RCA via median sternotomy. Cardiopulmonary bypass [CPB] was established, and the RCA was carefully dissected, ligated at its anomalous origin, and reimplanted onto the anterior surface of the ascending aorta. Intraoperative transesophageal echocardiography confirmed satisfactory flow through the reimplanted RCA, and the patient demonstrated an uneventful recovery. ***Conclusion:*** This case highlights the importance of recognizing high-risk coronary anomalies such as malignant RCA and the role of advanced imaging techniques in accurate diagnosis. Direct surgical translocation of the anomalous RCA is a safe and effective treatment option, providing a definitive solution that restores normal coronary anatomy and eliminates the risk of dynamic compression. Meticulous surgical technique, comprehensive postoperative monitoring and long-term follow-up are essential for optimizing patient outcomes.

Keywords: Anomalous right coronary artery, Interarterial course, Left coronary cusp, Surgical translocation, Myocardial ischemia, Coronary computed tomography angiography.

INTRODUCTION

Anomalous coronary artery origins are rare congenital anomalies, with an estimated prevalence ranging from 0.3% to 1.3% in the general population. Among these, the anomalous origin of the right coronary artery [RCA] from the left coronary cusp [LCC] with an interarterial course between the aorta and pulmonary artery is particularly concerning. This configuration, often termed a "malignant" RCA, is associated with an increased risk of myocardial ischemia, arrhythmias, and sudden cardiac death, especially during periods of heightened cardiac demand such as exercise.

The pathophysiological risk is primarily due to the dynamic compression of the anomalous RCA between the great vessels, compounded by high-risk anatomical features such as a slit-like ostium, acute take-off angle, and intramural course within the aortic wall.

These features can lead to compromised coronary blood flow, particularly during systole or tachycardia, resulting in transient myocardial ischemia. [1]

Clinical presentations of malignant RCA anomalies vary widely, from asymptomatic cases to exertional chest pain, syncope, palpitations, or even sudden cardiac death. The advent of advanced imaging modalities, particularly coronary computed tomography angiography [CCTA], has enhanced the detection and characterization of these anomalies, allowing for detailed assessment of coronary origin, course, and associated high-risk features. [2]

Management strategies for anomalous RCA with an interarterial course are guided by symptomatology, evidence of ischemia, and anatomical risk factors. Current guidelines recommend surgical intervention in symptomatic patients or those with high-risk anatomical features, even in the absence of symptoms. Surgical options include unroofing of the intramural segment, coronary artery bypass grafting [CABG], and direct reimplantation of the anomalous artery into the appropriate aortic sinus. Among these, direct reimplantation is considered a definitive approach that restores normal coronary anatomy and eliminates the risk of dynamic compression.

This case report details the successful surgical translocation of a malignant RCA originating from the LCC in a 38-year-old male presenting with recurrent angina, syncope, and palpitations. The report underscores the importance of accurate anatomical diagnosis, meticulous surgical planning, and the technical nuances of direct RCA translocation. We also discuss postoperative outcomes and the role of long-term follow-up in such patients.

CASE PRESENTATION

A 38-year-old male with underlying diabetes mellitus, hypertension, dyslipidemia, and a history of ischemic heart disease [IHD] presented with recurrent episodes of chest discomfort, occasional palpitations, and syncopal attacks, occurring approximately three times per month. He had a significant medical history of a motor vehicle accident in 2014, which required internal fixation for multiple fractures, including intramedullary nailing of the left femur, plating of the left humerus, and left tibia-fibula, followed by revision surgery for an infected femur implant.

Initial cardiovascular evaluation revealed a New York Heart Association [NYHA] functional class II and Canadian Cardiovascular Society [CCS] class I angina. Clinical examination showed stable vital signs and unremarkable systemic examinations. Resting electrocardiography [ECG] displayed sinus rhythm without ischemic changes.

A coronary computed tomography angiography [CTCA] performed for further evaluation revealed an anomalous right coronary artery [RCA] originating from the left coronary cusp [LCC] with an interarterial course between the aorta and pulmonary artery—an anatomical configuration classified as "malignant." Additionally, minimal calcification of the proximal left anterior descending artery [LAD] with a calcium score of 1 was noted. Further myocardial perfusion imaging showed mild inferior wall perfusion defect without inducible ischemia, and transthoracic echocardiography confirmed good left ventricular [LV] function with an ejection fraction of 68%.

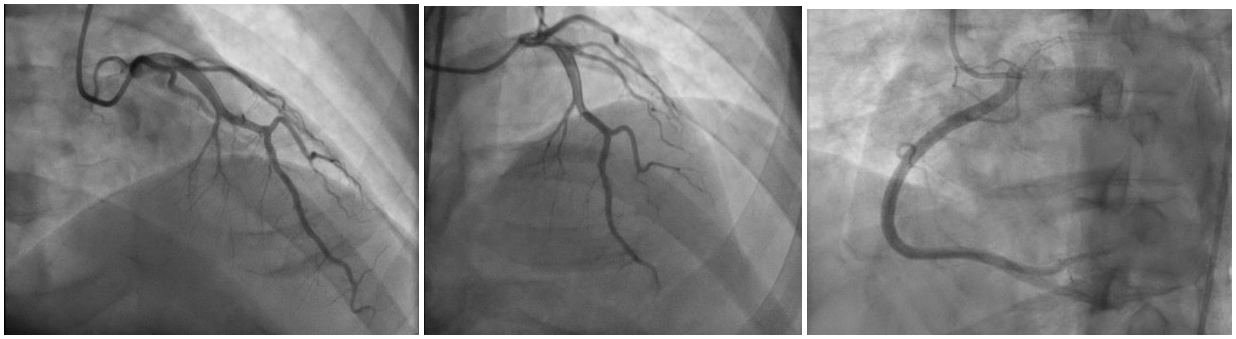

Figure 1: Coronary angiography revealed normal left coronary system anatomy, with unobstructed flow through the left anterior descending [LAD] and left circumflex [LCx] arteries. However, engagement of the right coronary artery [RCA] proved technically challenging. Multiple attempts with standard catheters failed to selectively cannulate the RCA from the right coronary sinus

Subsequent imaging and catheter maneuvers revealed that the RCA originated anomalously from the left coronary cusp [LCC], sharing a close proximity with the left main ostium. The anomalous origin and atypical take-off angle contributed to the difficulty in selective engagement, a hallmark feature often seen in cases of malignant RCA arising from the opposite sinus.

Due to the unusual origin and suspected interarterial course, a coronary CT angiogram [CTCA] was subsequently performed for detailed anatomical delineation.

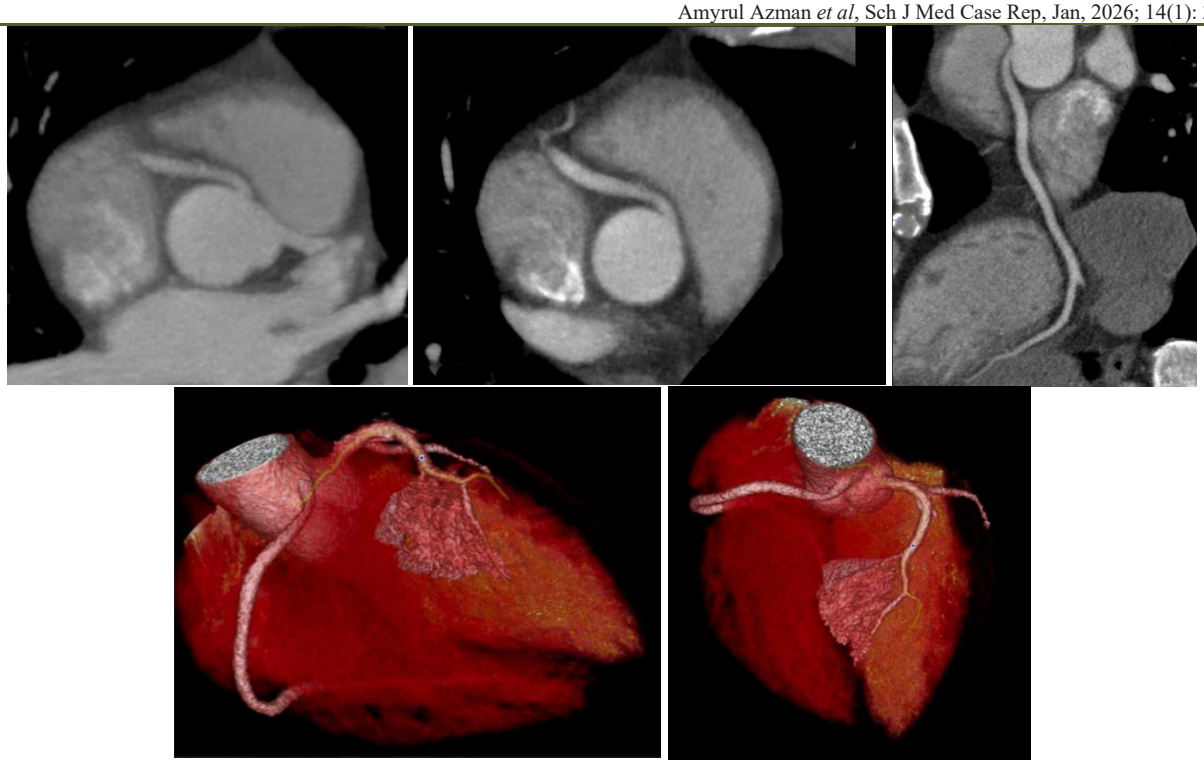

Figure 2: The right coronary artery [RCA] was found to arise anomalously from the left coronary sinus, adjacent to the origin of the left main coronary artery. It coursed between the aorta and pulmonary trunk [inter-arterial course], consistent with a malignant variant due to the risk of dynamic compression. The proximal RCA segment appeared slit-like and narrowed, suggesting ostial compromise, particularly during exertion

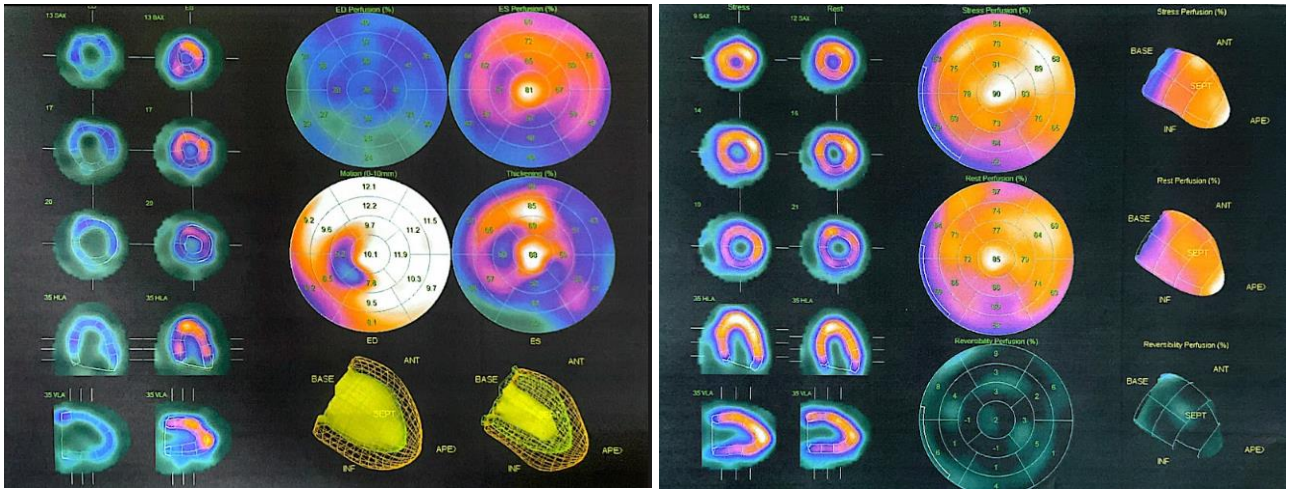

Figure 3: Rest–stress imaging demonstrated a moderate reversible perfusion defect involving the inferior and inferolateral walls, consistent with inducible ischemia in the RCA territory. The scan demonstrated inducible ischemia in the inferior wall corresponding to the anomalous RCA territory, reinforcing the need for definitive revascularization

The patient underwent median sternotomy, and the right internal mammary artery was harvested as a backup in case of any unforeseen complications. Following cardiopulmonary bypass [CPB] and aortic cross-clamping, the RCA was carefully dissected at its anomalous origin, ligated, and reimplanted onto the anterior aspect of the ascending aorta using a 7/0 Prolene suture. The procedure was completed with a total bypass time of 144 minutes and an aortic cross-clamp time of 95 minutes. Intraoperative transesophageal echocardiography confirmed good flow through the newly reimplanted RCA with excellent flow meter measurements post CPB.

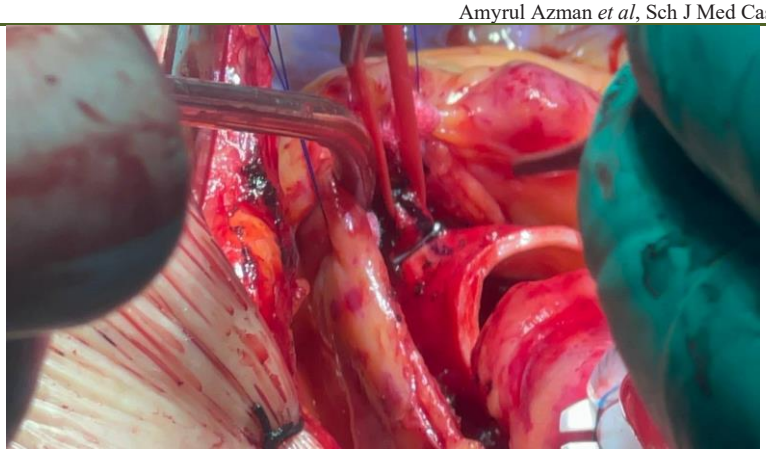

Figure 4: Pre-reimplantation - Intra-operative inspection following median sternotomy revealed an anomalous right coronary artery [RCA] originating from the left coronary sinus. The proximal RCA coursed intramurally and between the ascending aorta and pulmonary artery, consistent with a malignant interarterial course. The anomalous ostium was identified on the left side of the aortic root, and the proximal RCA appeared flattened and compressed at its origin

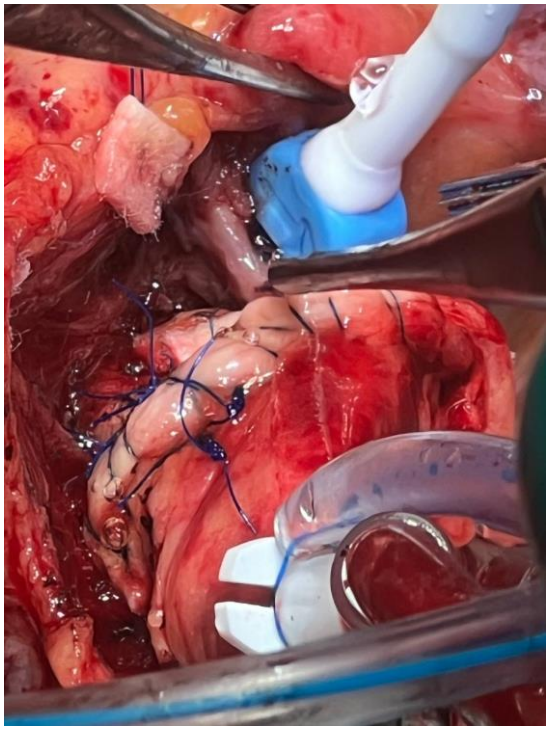

Figure 5: Post-reimplantation - Following cardiopulmonary bypass and transverse aortotomy, the anomalous RCA was ligated at its origin and mobilized. The vessel was then reimplanted into the appropriate right coronary sinus using an end-to-side anastomosis. Intra-operative assessment confirmed a tension-free anastomosis with good vessel alignment and restoration of a normal anatomical course, eliminating the interarterial compression

Postoperatively, the patient recovered well, with no evidence of myocardial ischemia or arrhythmias. He was discharged on day six with appropriate medical therapy, including beta-blockers, statins, and antiplatelets. At follow-up, the patient reported resolution of chest discomfort and palpitations, with stable LV function on echocardiography [EF 50%].

DISCUSSION

Coronary artery anomalies [CAAs] are a heterogeneous group of congenital coronary artery abnormalities that vary widely in their clinical significance. They are classified based on the origin, course, and termination of the coronary arteries. Among these, the anomalous origin of the right coronary artery [RCA] from the left coronary cusp [LCC] is a recognized subtype that can have significant clinical consequences, especially when it follows an interarterial course between the aorta and pulmonary artery.

A malignant right coronary artery [RCA] with an interarterial course between the aorta and pulmonary artery poses a risk of dynamic compression, especially during exertion. Pathophysiologic features such as a slit-like ostium, acute take-off angle, and intramural segments contribute to transient ischemia and potentially sudden cardiac death. Diagnosis requires a multimodal

approach, with coronary CT angiography [CTCA] as the gold standard for anatomical assessment. Other tools include ECG, stress testing, angiography, and cardiac MRI.

Surgical treatment is indicated for symptomatic or high-risk patients. Options include unroofing, CABG, reimplantation, and direct translocation. In this case, direct RCA translocation to the ascending aorta was performed due to favorable anatomy and absence of diffuse coronary disease. This method restored normal coronary physiology and eliminated the risk of dynamic compression.

A "malignant" course typically refers to the interarterial path between the aorta and pulmonary artery, which can be further complicated by an intramural segment within the aortic wall. This anatomy predisposes the patient to coronary compression during exertion, leading to myocardial ischemia or fatal arrhythmias. [6] The authors stress the importance of multimodality imaging. Coronary computed tomography angiography [CCTA] is highlighted as the most accurate non-invasive modality for delineating the course and origin of the anomalous artery. Functional imaging, such as stress cardiac MRI or nuclear perfusion imaging, is useful for detecting inducible ischemia. [6]

The management of anomalous right coronary artery [RCA] with an interarterial course—often referred to as "malignant RCA"—requires a carefully selected surgical approach to ensure optimal patient outcomes. Both the technique described by O'Connell and Tendulkar [2021] and the approach utilized in this case report focus on direct reimplantation of the RCA, but they differ significantly in their technical execution.

O'Connell and Tendulkar [2021] introduced a novel method involving direct anatomic reimplantation of the anomalous RCA without performing an aortotomy. In this approach, the RCA was dissected, mobilized from its anomalous origin, and directly reimplanted into an appropriate position on the ascending aorta. By avoiding an aortotomy, the authors minimized the complexity of the procedure, reduced the risk of aortic wall injury, and avoided the potential complications associated with aortic closure. This technique is particularly advantageous in terms of reducing operative time and minimizing bleeding risks. Furthermore, the direct reimplantation without aortotomy allows for a more streamlined surgical process while maintaining the anatomical and physiological integrity of the coronary artery.

In contrast, the technique utilized in the current case report involved a median sternotomy with aortic exposure and controlled aortotomy. This method provided direct visualization of the aortic root and coronary ostia, enabling precise identification of the anomalous RCA origin. The RCA was carefully dissected, ligated at its original site, and translocated to a newly created ostium on the anterior ascending aorta. This approach, while technically more complex due to the need for aortotomy, offered several key advantages. The direct visualization and exposure allowed for meticulous anastomosis, ensuring accurate alignment and secure reimplantation of the RCA. Moreover, it allowed for the closure of the original anomalous RCA origin with direct suture, reducing the risk of residual blood flow or pseudoaneurysm formation. The use of an aortotomy also enabled efficient management of any unexpected anatomical variations encountered during the procedure.

While both techniques aim to restore normal coronary physiology and eliminate the risk of dynamic compression, each has its own strengths and limitations. The non-aortotomy technique described by O'Connell and Tendulkar is less invasive, faster, and may be preferable in patients with fragile aortic tissue or when minimizing operative time is critical. However, it may lack the precision and control offered by the aortotomy approach. Conversely, the aortotomy-based method, as demonstrated in this case, provides superior visualization and control, but at the cost of a slightly more complex procedure and a potentially higher risk of aortic complications.

In clinical practice, the choice between these two techniques should be guided by the patient's anatomy, the surgeon's expertise, and intraoperative findings. Understanding the advantages and limitations of each approach allows for tailored surgical planning, ensuring the best possible outcomes for patients with malignant RCA. [3]

In their comprehensive review, Bigler *et al.,* [2022] discuss the current understanding, challenges, and therapeutic strategies in managing anomalous coronary arteries originating from the opposite sinus of Valsalva [ACAOS]—particularly those with an interarterial and intramural course, such as a malignant right coronary artery [RCA] arising from the left coronary cusp [LCC]. The authors emphasize the clinical ambiguity and diagnostic complexity surrounding ACAOS, especially in asymptomatic individuals or those presenting with nonspecific symptoms like chest pain or syncope. [4] Key takeaways include the importance of morphological assessment using multi-modality imaging—such as CT coronary angiography and cardiac MRI—to determine the high-risk features of ACAOS. These features include an interarterial course, intramural segment, slit-like ostium, and acute take-off angle, which collectively may predispose to myocardial ischemia or sudden cardiac death [SCD], particularly during exertion.

Bigler *et al.,* highlight a stepwise and individualized management approach based on patient symptoms, ischemia evidence, and anatomical risk markers. They note that in symptomatic patients or those

with documented myocardial ischemia, surgical correction remains the mainstay of therapy. The three primary surgical options outlined include:

a) Unroofing of the intramural segment [typically favored for intramural left coronary arteries],
b) Pulmonary artery translocation to decompress the RCA,
c) Coronary reimplantation [translocation]—particularly indicated for anatomies where intramural unroofing is not feasible or where a more physiological correction is preferred. [4]

In another detailed review, Gharibeh *et al.,* [2021] analyze and compare the various surgical strategies available for correcting anomalous origin of the right coronary artery [RCA] from the left coronary sinus, particularly when it courses between the aorta and pulmonary artery—an anatomy associated with a heightened risk of myocardial ischemia and sudden cardiac death [SCD]. [5] The authors underscore that surgical correction is indicated in symptomatic patients or those with ischemia on testing. Four main surgical approaches are reviewed:

a) Unroofing of the intramural segment: Often considered first-line for intramural variants; involves opening the aortic wall overlying the intramural RCA to create a new ostium. While this is technically straightforward, risks include aortic insufficiency and damage to the aortic valve commissure.
b) Pulmonary artery translocation: Involves moving the pulmonary artery away from the anomalous RCA to relieve external compression. This method is less favored due to limited long-term data and potential for recurrence of symptoms.
c) Osteoplasty: Aimed at enlarging the native slit-like RCA ostium. Although less invasive, it may not adequately resolve flow restriction or eliminate the interarterial compression.
d) Anatomic reimplantation [translocation]: Considered the most definitive and anatomically restorative approach, this technique entails transecting the anomalous RCA and reimplanting it directly into the appropriate aortic sinus. It provides a physiological origin and course, eliminating the risks of external compression or kinking. However, it requires meticulous surgical planning and is more technically demanding.

The review concludes that patient-specific anatomy, presence of intramural course, and surgical expertise should guide the choice of intervention. The authors favor anatomic reimplantation, especially in young, active patients and when complete correction with low risk of recurrence is desired. [5]

Raikar *et al.,* [2022] present a detailed case study of an elderly female in her late 70s who presented with exertional syncope—a rare but serious manifestation of an anomalous right coronary artery [RCA] arising from the left coronary cusp [LCC], following a malignant interarterial course between the aorta and pulmonary artery. [7] Despite her high-risk anatomy, the patient opted for conservative management due to her age and comorbidities. The authors highlight that although surgical options—including unroofing, reimplantation, bypass grafting, and osteoplasty—are recommended for young symptomatic patients or those with ischemia, conservative treatment with activity restriction may be acceptable for older or asymptomatic individuals. The present case, involving a 38-year-old male with exertional symptoms and a confirmed malignant course of the right coronary artery [RCA] arising from the left coronary cusp, closely aligns with the therapeutic recommendations outlined by Raikar *et al.,* [2022]. While their case described an elderly female managed conservatively due to age and comorbidities, the decision to proceed with surgical translocation in our patient reflects an individualized, anatomy- and age-guided approach. Given the high-risk anatomical features—namely the interarterial course and symptom burden—early surgical correction was deemed both appropriate and necessary to mitigate the risk of future ischemic events or sudden cardiac death. This case reinforces the growing consensus that younger symptomatic patients with malignant RCA variants benefit most from timely surgical intervention.

The surgical technique employed in our case closely mirrors the structured approach described in the Multimedia Manual of Cardiothoracic Surgery [MMCTS] tutorial on the correction of anomalous right coronary artery [RCA] from the left coronary sinus. The tutorial outlines the rationale and step-by-step method for direct RCA reimplantation, particularly in patients with a malignant interarterial course, as seen in our 38-year-old symptomatic male patient. Our decision to proceed with median sternotomy, RCA mobilization, ligation at the anomalous origin, and direct anastomosis to the anterior ascending aorta aligns with the MMCTS technique. This approach ensures restoration of a physiological coronary course while mitigating the risks of dynamic compression between the aorta and pulmonary artery. Additionally, the tutorial's emphasis on careful anastomotic tension, intraoperative flow verification, and postoperative imaging aligns with the meticulous intraoperative and recovery strategies applied in our case. [8] As demonstrated, this surgical method offers a durable and anatomically sound solution for high-risk anomalous coronary anatomy, with favorable short- and long-term outcomes.

The case described by Irshad *et al.,* [2024] closely parallels our patient's presentation, reinforcing the clinical significance of malignant RCA anomalies arising from the left coronary cusp with an interarterial and intramural course. Both patients were symptomatic—ours with exertional syncope and the

reported case with positional angina—highlighting the variability in symptomatology that can occur with similar underlying anatomy. Importantly, both cases underline the limitations of conventional coronary angiography in detecting anomalous RCA origins, emphasizing the diagnostic value of CT coronary angiography [CTCA] in identifying high-risk features such as slit-like ostia, intramural segments, and interarterial trajectories. While Irshad *et al.,* managed their patient with coronary artery bypass grafting [CABG] to the mid-RCA, our approach involved direct surgical reimplantation of the RCA onto the anterior ascending aorta after ligating its anomalous origin. [9] This distinction reflects the spectrum of accepted surgical strategies for malignant RCA anomalies. Ultimately, both cases demonstrate that when high-risk anatomical features and symptomatic burden are present in relatively young patients, early surgical correction—tailored to anatomical feasibility—offers a definitive solution to prevent ischemic complications and potential sudden cardiac death.

This case underscores the importance of a comprehensive diagnostic approach, careful surgical planning, and individualized patient management. Recognition of high-risk coronary anomalies and timely surgical intervention can significantly improve patient outcomes, transforming a potentially fatal condition into a manageable one.

CONCLUSION

This case report highlights the successful surgical management of a malignant right coronary artery [RCA] originating from the left coronary cusp [LCC] with an interarterial course between the aorta and pulmonary artery—a high-risk congenital coronary anomaly associated with significant morbidity and potential mortality. The patient's presentation with recurrent angina, palpitations, and syncope underscored the functional impact of the anomalous RCA, despite the absence of significant obstructive coronary artery disease on conventional angiography.

The decision to pursue surgical intervention was guided by a comprehensive assessment, including advanced imaging techniques such as coronary computed tomography angiography [CTCA] and myocardial perfusion imaging, which confirmed the high-risk anatomy of the RCA. Direct translocation of the RCA to a safe location on the ascending aorta was chosen as the definitive surgical strategy. This approach was favored over other options such as unroofing or coronary artery bypass grafting [CABG] due to its ability to restore normal coronary anatomy and ensure unrestricted coronary blood flow without the risks associated with graft patency.

The surgical procedure was performed with careful dissection, precise anastomosis, and meticulous intraoperative monitoring, resulting in a successful outcome. The patient experienced a smooth postoperative recovery with no evidence of ischemic complications, arrhythmias, or other adverse events. Long-term follow-up demonstrated complete resolution of symptoms, with stable left ventricular function and patent reimplanted RCA on imaging.

This case underscores several critical learning points:

Importance of Early Diagnosis: A high index of suspicion is required for malignant coronary anomalies, especially in patients with recurrent angina, syncope, or palpitations, even in the absence of significant coronary artery stenosis on conventional angiography.

Role of Advanced Imaging: CTCA is invaluable for accurately characterizing coronary artery anatomy, identifying high-risk features, and guiding clinical decision-making.

Surgical Precision Matters: Direct translocation or reimplantation of a malignant RCA offers a definitive solution, but requires meticulous surgical technique to ensure long-term patency and avoid complications.

Postoperative Vigilance: Regular follow-up is essential to monitor for recurrent symptoms, assess left ventricular function, and ensure long-term patency of the reimplanted coronary artery.

In conclusion, this case demonstrates that surgical translocation of a malignant RCA is a safe and effective treatment option for patients with symptomatic or high-risk anatomical variants. Early diagnosis, individualized surgical planning, and comprehensive postoperative follow-up are critical for optimizing patient outcomes.

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