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A Rare Case of Giant Aneurysm of the Right Coronary Artery with Right Coronary Artery to Coronary Sinus Fistula

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Abstract

Background: Coronary artery aneurysms are rare, localized, abnormal dilations of the artery. The combination of coronary artery aneurysm and coronary artery to coronary sinus fistula is even more rare. **Case Presentation:** This report presents a case of a giant right coronary artery aneurysm and a right coronary artery to coronary sinus fistula in a 55-year-old Indian female who presented with chest pain and exertional dyspnea that had been increasing over a six-month period and became severe one week prior to admission. The referral diagnosis was severe aortic regurgitation with a ruptured sinus of Valsalva; however, diagnostic imaging at our center revealed a 100 mm aneurysm of the proximal to mid segment of the right coronary artery with ectasia of the distal right coronary artery and a fistulous connection **at the level of its ostium.** The patient underwent successful aneurysm resection, fistula closure and coronary artery bypass grafting using great saphenous vein graft to the posterior descending artery. **Conclusions:** This case highlights the importance of including giant coronary artery aneurysms in the differential diagnosis for patients presenting with chest pain, exertional dyspnea, and a cardiac mass. It also emphasizes the need to investigate for coronary fistulae as a significant associated finding and underscores the diagnostic challenges posed by this complex pathology.

Subject Areas

Cardiology, Surgery & Surgical Specialties

Keywords

Giant Coronary Artery Aneurysm, Coronary Sinus Fistula, Case Report

1. Introduction

Coronary artery aneurysms (CAAs) are rare, localized, abnormal dilations of an artery, defined as an increase in diameter of 1.5 to 2 times that of an adjacent normal vessel segment, with an estimated incidence of 1.5% - 2.0% in pre- and post-mortem studies [1] [2]. Giant coronary artery aneurysms (defined as those with a diameter greater than 40 mm) are even rarer, with a reported prevalence of 0.02% to 0.2% [1].

Coronary arteriovenous fistula is a rare congenital anomaly presenting in later life as an incidental finding or with major potential complications such as congestive heart failure, myocardial infarction, thrombosis, endocarditis, arrhythmia and even sudden death. The degree of blood shunting and its consequent hemodynamic impact are determined by the size of the fistula and the pressure gradient between the coronary artery and the receiving cardiac chamber [2]. The flow from the fistula increases flow circulating through the coronary, potentially enhancing the capacity for aneurysmal growth.

This case report describes a patient presenting with a combined giant RCA aneurysm and an RCA to CS fistula, a rare anatomical variant with limited representation in the current literature. This unusual presentation necessitates a comprehensive review of the existing literature to understand the etiology, clinical presentation, diagnostic approaches, and management strategies for such complex coronary anomalies.

2. Case Presentation

A 55-year-old Indian female was referred to our institution for surgical management with the diagnosis of severe aortic regurgitation and ruptured sinus of Valsalva. She presented with worsening chest pain and exertional dyspnea. Her symptoms had been present for six months, with progressive worsening, becoming severe one week prior to admission. She did not have a history of trauma nor previous invasive cardiac interventions. No history of hypertension, chronic infections, diabetes mellitus and no family history of aneurysms. At presentation, she was hemodynamically stable but had orthopnea. No murmur of aortic regurgitation was detected upon auscultation, and no other significant findings were noted on clinical examination.

The baseline complete blood count (CBC) showed moderate normocytic, normochromic anemia but normal white cell counts. The renal function tests, thyroid function tests, coagulation profile and lipid profile were all normal.

The findings of aortic regurgitation and a ruptured sinus of Valsalva (RSOV) were from a transthoracic Echocardiogram from the referring center. The left ventricular systolic function was reported to be normal on the same echocardiogram. Coronary angiography performed revealed findings consistent with severe coronary artery disease (total occlusion of the mid-RCA). **We suspect that the massive volume of the aneurysm, combined with rapid contrast run-off into the low-pressure coronary sinus, resulted in severe contrast dilution. This hemo-**

dynamic alteration could have prevented vessel opacification during angiography, mimicking a total occlusion.

However, Cardiac CT angiography at our institution showed features of a giant fusiform RCA aneurysm with an ectatic and tortuous RCA segment distal to the aneurysm (Figures 1-3) and an RCA to coronary sinus fistula **at the level of the ostium** (Figure 4).

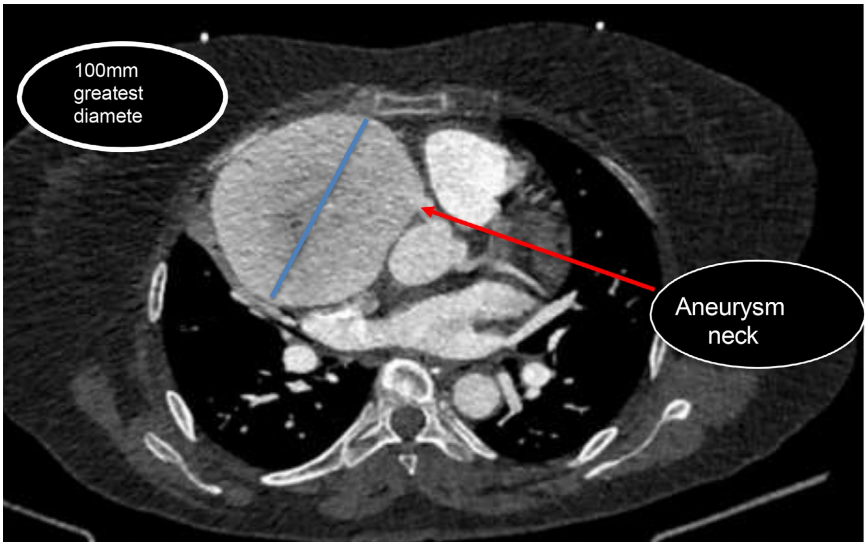


Figure 1. CT angiogram (axial view) showing large/giant RCA aneurysm measuring 100 mm in greatest diameter (blue line), red arrow shows neck of the aneurysm.



Figure 2. CT. angiogram showing Ectatic and tortuous RCA distal to the aneurysm.

The patient underwent surgical resection of the RCA aneurysm, fistula closure and coronary artery bypass grafting (CABG) using a saphenous vein graft (SVG)

from proximal ascending aorta to Posterior descending artery (PDA) (Aorta-SVG-PDA graft).



Figure 3. CT. angiogram (coronal view) showing tortuous distal RCA.

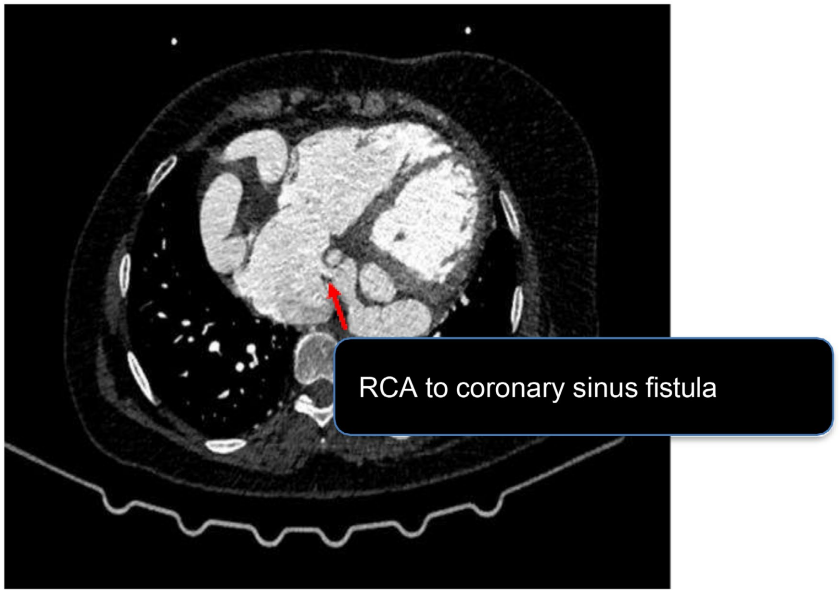


Figure 4. CT angiogram (axial view) showing Distal RCA to Coronary sinus fistula (red arrow).

A median sternotomy was performed, revealing a discrete aneurysm within the proximal-mid segment of the right coronary artery (RCA), situated anterior to the right atrium (RA) (**Figure 5(A)**). Distal to the aneurysmal dilatation, the RCA exhibited ectasia and tortuosity, with a fistula connecting the RCA to the coronary

sinus (CS) (Figure 5(B), Figure 5(C)). No significant collateral circulation was identified. Furthermore, no evidence of a ruptured sinus of Valsalva was observed.

Cardiopulmonary bypass (CPB) was initiated via aorto-bicaval cannulation. Following the application of an aortic cross-clamp, a transverse aortotomy was performed. Cardiac arrest was induced by the antegrade administration of cardioplegia directly into the left main coronary ostium.

Upon opening the giant RCA aneurysm, it was found to be free of thrombus and perforators. The RCA ostium was noted to open directly into the aneurysm. Cardioplegia was then directly administered into the distal RCA. The RCA ostium was subsequently closed with a pericardial patch. Examination of the aortic valve revealed no abnormalities.

The aneurysmal and ectatic segments of the RCA, extending to the crux, were resected. The fistula into the coronary sinus (at the level of the ostium) was closed using a 7/0 Prolene transfixion suture without opening into the right atrium. The distal RCA was transfixed at the level of the crux, and the aortotomy was closed with a 4/0 Prolene suture.

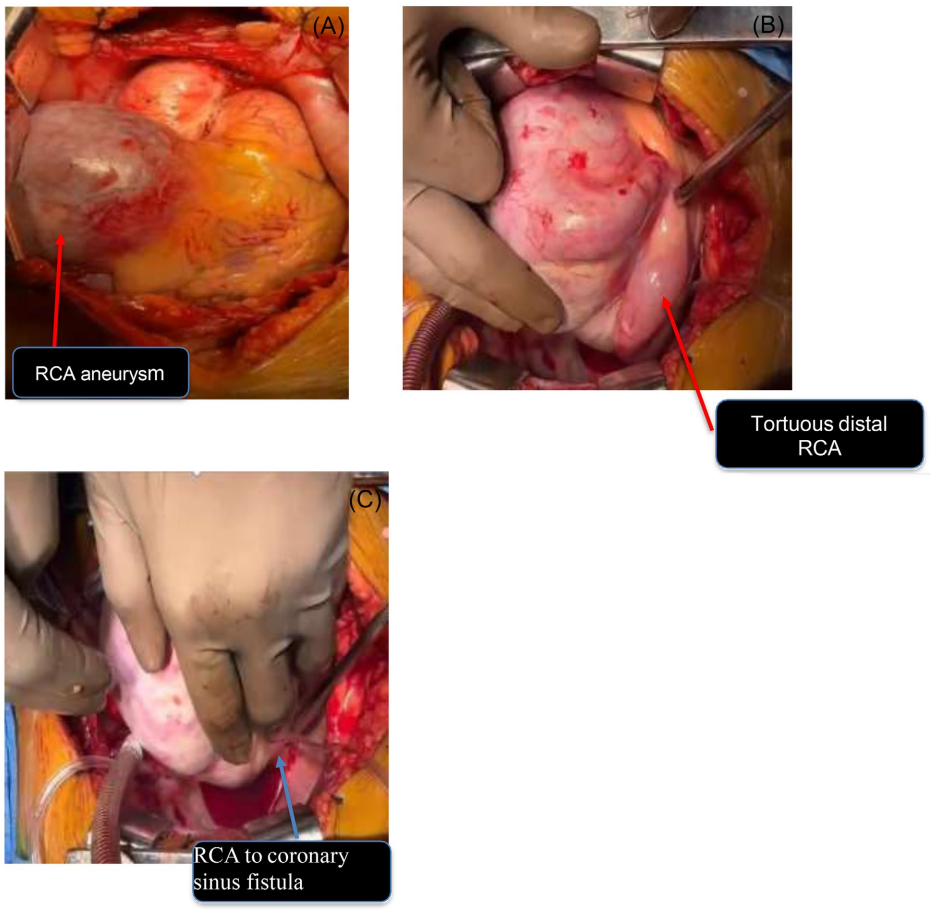


Figure 5. (A-C): Intra-operative images showing the giant RCA aneurysm (red arrow in Figure 5(A)). RCA distal to the aneurysm was ectatic and tortuous (red arrow in Figure 5(B)). distally, the RCA to CS fistula is shown by the blue arrow in Figure 5(C).

Coronary artery bypass grafting (CABG) was performed utilizing a saphenous vein graft (SVG) anastomosed to the posterior descending artery (PDA) with 7/0 Prolene sutures for both proximal and distal anastomoses (Figure 6).

The patient experienced an uneventful postoperative recovery. Postoperative echocardiography confirmed the successful closure of the fistula and demonstrated normal left ventricular systolic function. The patient was discharged on the fifth postoperative day **on a dual antiplatelet therapy regimen consisting of Aspirin 75 mg and Clopidogrel 75 mg daily**. At the one-month follow-up, the patient remained asymptomatic and in good health. **The planned surveillance strategy included a CT coronary angiography scheduled at 6 months and 1 year postoperatively to monitor graft patency and ensure no aneurysmal recurrence.**

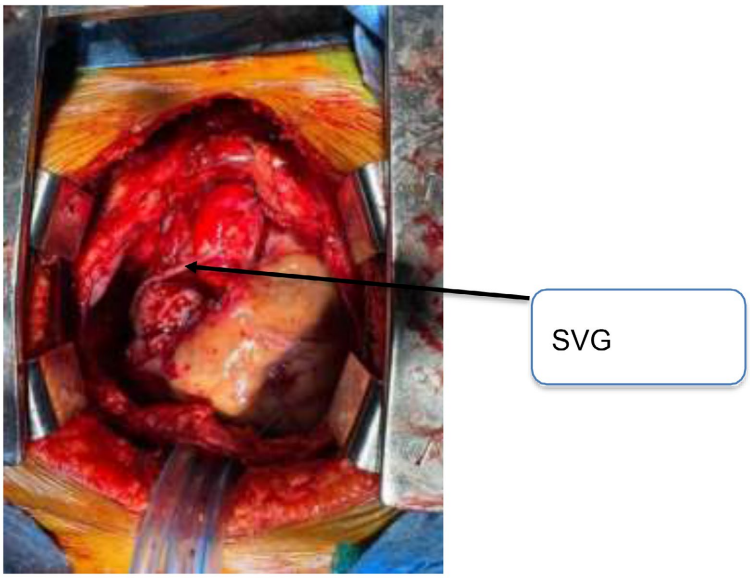


Figure 6. Intraoperative image showing the bypass graft after aneurysm resection (Aorta-SVG-PDA graft), black arrow.

Patient perspective;

"*After enduring six months of worsening chest pain and shortness of breath that made even lying down difficult, I was anxious about undergoing major heart surgery. However, the recovery process was much smoother than I anticipated. I woke up feeling relieved, and my progress was steady enough that I was able to leave the hospital just five days after the operation.*"

3. Discussion

The definition of a "giant" coronary artery aneurysm lacks precision, particularly when a coronary fistula is also present. Of cases that have been reported as a "giant" coronary aneurysm with coronary fistula, 70% were >30 mm in diameter; and 68% were in women. With regard to location, the RCA was the main vessel affected (involved in 40.5% of cases) [3].

In combined coronary artery aneurysm and fistula connection, the draining site of the fistula is commonly the pulmonary artery, the right atrium, and the left ventricle. The pathophysiology of aneurysm formation in the presence of a coronary fistula is thought to involve chronic volume overload and increased shear stress within the affected artery. The persistent runoff into a low- pressure chamber, in this case, the coronary sinus, may promote progressive dilation of the vessel wall.

The largest reported RCA aneurysm measured 160 mm in diameter but had a fistula to the left ventricle [4]. **Based on a comprehensive literature search of PubMed and Google Scholar databases utilizing the terms 'giant right coronary artery aneurysm' and 'coronary sinus fistula',** an RCA aneurysm with a fistula draining into the coronary sinus has been described in only five previous case reports [5]-[9] as summarized in **Table 1** below.

Table 1. Summary of published case reports of RCA aneurysms combined with RCA to coronary sinus fistula.

Author	Year	Journal	Diameter of RCA aneurysm	Site of fistula connection
Shahid *et al.*	2025	*World Journal of Clinical Surgery*	56 × 58 mm	Coronary sinus
Awad *et al.*	2025	*JACC Case Reports*	20 × 25 mm	Coronary sinus
Yamazaki *et al.*	2025	*European Heart Journal - Case Reports*	10 × 16 mm	Coronary sinus
Briosa e Gala *et al.*	2021	*Revista Portuguesa de Cardiologia*	58 × 56 mm	Coronary sinus
Komoda *et al.*	2010	*General Thoracic and Cardiovascular Surgery*	40 mm	Coronary sinus

This case report therefore documents the largest RCA aneurysm (of 100 mm) that drains into the coronary sinus, a rare occurrence. Our patient's pathology parallels a previously reported 10.6 × 9.2 cm giant right coronary aneurysm [10] in terms of location and magnitude, but diverges clinically by presenting with cardiac failure symptoms secondary to a fistula, rather than only the compressive sequelae observed in their report.

While coronary artery aneurysms are most commonly caused by atherosclerotic coronary artery disease, other etiologies include congenital malformations, connective tissue disorders, infections, and Kawasaki disease. Congenital aneurysms are typically larger than those of atherosclerotic origin and are most commonly located in the right coronary artery [3] [9]. Reported complications of these aneurysms include thrombosis and distal embolization, rupture, and vasospasm.

Most coronary fistulas are also congenital in origin, resulting from the persistence of junctions of primordial epicardial vessels with intramyocardial sinusoidal circulation during embryonic development [11] but non-congenital fistulas (e.g., iatrogenic fistulas, a consequence of chest trauma) have been reported.

Considering the large size of this aneurysm and the presence of a draining fistula, this anomaly was presumed to be of congenital origin. The absence of thrombus or perforators within the aneurysm sac in our case suggests a relatively lami-

nar flow pattern, possibly contributing to the aneurysm's silent expansion over time. Atherosclerotic disease was considered unlikely due to the larger size of the aneurysm, presence of a fistula connection, patient's normal lipid profile and the absence of other cardiovascular risk factors like obesity, hypertension and diabetes. Furthermore, vasculitis, infectious etiologies, and connective tissue diseases were much more unlikely given the normal infection and inflammatory makers, and the absence of any syndromic features on clinical examination.

This case also underscores the diagnostic limitations of transthoracic echocardiography in evaluating complex coronary anomalies. The initial misdiagnosis of a ruptured sinus of Valsalva highlights the need for high-resolution imaging when clinical suspicion persists. Cardiac CT angiography provided superior anatomical delineation, revealing the true pathology and guiding surgical planning. In similar cases, multimodal imaging-including transesophageal echocardiography and cardiac MRI, may be warranted to fully characterize the lesion and its hemodynamic impact.

Surgical intervention remains indicated for symptomatic patients with symptomatic obstructive coronary artery disease and site-specific targets such as large CAAs at the bifurcation of the left main, giant coronary aneurysms that are at risk of rupture; and those CAAs associated with hemodynamically significant fistulas. There are no guidelines to help clinicians as it relates to size in asymptomatic patients. One might extrapolate from the aortic literature, but this approach is limited, and we rely on anecdotal recommendations to intervene when the diameter exceeds 3 - 4 cm or when growth is identified with serial imaging [12].

A coronary aneurysm with a diameter three to four times that of the native vessel is considered an absolute indication for surgical intervention to mitigate the risk of rupture, which increases at diameters greater than 30 mm. Surgical options include: resection of the aneurysm with CABG, suture closure of the proximal and distal ends of the aneurysm, closure with a woven Dacron patch, and aneurysmal plication. The surgical management of coronary fistulas includes obliteration by double ligation of the vessel. Some surgeons have used tangential arteriorrhaphy, others have performed an arteriotomy and sutured the fistulous stoma [13]. The giant nature and tortuosity of the aneurysms associated with fistulas restrict catheter interventions.

For this patient, surgical intervention was indicated due to the aneurysm's size (100 mm), the presence of a hemodynamically significant fistula, the risk of rupture and presence of symptoms. The decision to perform aneurysm resection with coronary artery bypass grafting (CABG) was based on the aneurysm's location, the absence of suitable distal vessel for reconstruction, and the need to eliminate the fistulous connection. Alternative techniques such as aneurysmal plication or patch repair were deemed less favorable due to the vessel's tortuosity and ectasia. The fistula was closed using a transfixion suture at the coronary artery to coronary sinus junction, avoiding entry into the right atrium and minimizing the risk of arrhythmia or bleeding.

Postoperatively, the patient had an uneventful recovery. Long-term surveillance with serial imaging is essential to monitor graft patency. Ultrafast computed tomography (UFCT) and echocardiography are used for follow-up examination [13].

Although no formal guidelines exist for postoperative management in such cases, antiplatelet therapy is typically continued to maintain graft flow and reduce thrombotic risk.

This case contributes to the limited literature on RCA aneurysms with RCA to coronary sinus fistulae and highlights the importance of considering rare coronary anomalies in patients with unexplained chest pain, exertional dyspnea, or cardiac masses.

4. Conclusion

We have reported successful surgery for a rare case of giant RCA aneurysm with RCA to CS fistula. This case highlights the importance of considering giant CAAs in the differential diagnosis of a patient with chest pain, exertional dyspnea and a cardiac mass and emphasizes the need to screen for coronary fistulae as a significant association. It also highlights the diagnostic challenges that may arise with such complex and rare cases.

Ethics Approval

We obtained ethical approval from the Institutional Ethics Committee- Yashoda academy of medical education and research (IEC-YAMER) with reference number: CR/PP/HTC-03/2025. All methods were performed in accordance with the Declaration of Helsinki.

Consent for Publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor in-Chief of this journal.

Consent to Participate

Informed consent to participate was obtained from the patient prior to inclusion in this case report. The patient understood that her anonymized clinical information would be used for academic and scientific purposes and freely agreed to participate in the study.

Availability of Data and Materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

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Author Contributions

K.A was the primary/corresponding author. Pioneered the case report writing and publication.

H.A helped in proofreading of the case report.

V.A helped to get patient consent and ethical approval for the case report.

Conflicts of Interest

The authors declare that they have no competing interests.

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List of Abbreviations

RCA: Right Coronary Artery

CAAs: Coronary Artery Aneurysms

CS: Coronary Sinus

UHI: Uganda Heart Institute

RSOV: Ruptured Sinus of Valsalva

CT: Computed Tomography

CBC: Complete Blood Count

RA: Right Atrium

CPB: Cardiopulmonary Bypass

CABG: Coronary Artery Bypass Grafting

SVG: Saphenous Vein Graft

PDA: Posterior Descending Artery

IEC-YAMER: Institutional Ethics Committee-Yashoda Academy of Medical Education and Research