

CASE REPORT

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Diagnosis and demonstration of single coronary artery by multidetector CT angiography: series of two cases

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Abstract

Background: Single coronary artery (SCA) is a rare anomaly, usually diagnosed incidentally during coronary artery angiogram. Individuals with this anomaly mostly remain asymptomatic while some present with symptoms such as chest pain, dyspnoea and even sudden death. The origin and the proximal course of anomalous coronary artery are the main prognostic factors. Several classification systems have been proposed based on the site of origin and anatomical distribution of anomalous artery. Coronary computed tomography angiography has become the reference method for such an assessment noninvasively. Herein, we report a series of two cases of SCA diagnosed on CT angiography. In one case it was single left coronary artery associated with other congenital cardiac anomalies, whereas in other it was single right coronary artery and was an isolated anomaly.

Case presentation: Our first case was of a 19-year-old female who presented with chest pain and dyspnoea. Transthoracic Echocardiography (TTE) features were suggestive of Tetralogy of Fallot (TOF) with infective endocarditis. Cardiac CT angiography revealed the presence of a large SCA arising from left aortic sinus with absence of normal origin of right coronary artery (RCA). This artery was dividing into and supplying different coronary arterial territories with pre-pulmonic course of RCA. The patient underwent Aortic valve replacement with pulmonary Commissurotomy and improved in post-operative period. Our second case was of a 50-year-old man with complaints of breathlessness and normal ECG and Echocardiography examination findings. Coronary CT angiography revealed the presence of SCA arising from right aortic sinus and supplying the territories of both RCA and Left coronary artery (LCA). The patient was managed conservatively with emphasis on aggressive control of risk factors.

Conclusions: SCA is a rare anomaly and may lead to catastrophic life threatening complications. The accurate delineation of the origin and course of the anomalous vessel is of paramount importance while planning surgical intervention. Management usually involves a multi-disciplinary approach with cardiologists and cardiac surgeons aiming for deciding an individual plan based on presentation and anatomy of each case.

Keywords: Single coronary artery, Anomalous coronary artery, Coronary artery anomalies, Lipton classification, CT angiography

Background

Coronary artery anomalies (CAA) comprise a wide group of rare congenital malformations, some of them remain asymptomatic while the others present with symptoms such as chest pain, breathlessness and even sudden death. A single coronary artery (SCA) is a rare anomaly which is defined as single coronary artery arising from the aortic trunk by a single coronary sinus

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and providing for perfusion of the entire myocardium regardless of its distribution [1]. SCA can be either an isolated anomaly or associated with other congenital abnormalities such as coronary artery fistula, bicuspid aortic valve, transposition of great vessels, coronary arterio-venous fistula, tetralogy of Fallot (TOF), truncus arteriosus, inter-ventricular septal defect, patent ductus arteriosus and patent foramen ovale [2].

SCA is usually an incidental finding with an estimated incidence ranging between 0.024% and 0.066% among patients undergoing coronary artery catheterization for various reasons [2]. Its prevalence among patients undergoing coronary computed tomography angiography (CTA) ranges between 0.024 and 0.098% [3–6]. In spite of the fact that conventional coronary angiography is the gold standard procedure for assessment of coronary arteries, it has disadvantages of being an invasive procedure with limited ability to display the complex anatomy in cases of CAA [7]. In contrast, CTA allows accurate and non-invasive depiction of such anomalies due to its multiplanar reformation capabilities [8].

Different classification systems of SCA based on necropsy findings and conventional coronary angiography exist in the published literature [9, 10]. Lipton et al. classified SCA based on the site of origin and anatomical distribution into two main categories: “R” right type and “L” left type. These two types were further subdivided into I, II, and III groups according to the anatomical course of the branches. In group I the solitary vessel follows the course of either a normal right or left coronary artery (R-I or L-I, respectively) with a continuation into the missing artery’s territory.

In group II, a SCA arises from right or left coronary sinus, and from its proximal segment, a large aberrant trunk arises that crosses the base of the heart to reach in the vicinity of a normally coursing contralateral coronary artery. The aberrant trunk can follow any one of the several different pathways to get to its vascular territory. These pathways are as follows: type A (anterior to the right ventricular conus or pulmonary artery); type B (between the aorta and pulmonary trunk); type P (posterior to aortic root); type S (above the inter-ventricular septum); type C (combined type). In type III, the LCX and LAD have separate origins from the proximal RCA [10, 11]. Only a limited number of studies have been published in the literature. Herein, we report a series of two cases of single coronary artery diagnosed on CTA. Our first case was a single L-II A type of SCA anomaly associated with other congenital cardiac anomalies. Our second case can be classified as an isolated R-I type of SCA anomaly.

Case presentation

Case 1

A 19-year-old female presented with chief complaint of on and off chest pain for 1 year and dyspnoea on exertion for the last 2 months. On examination, the patient was cyanotic and had tachycardia (HR-92 beats per minute) and tachypnea. TTE was done for cardiac evaluation and revealed severe right ventricular outflow tract obstruction with infundibular stenosis. A small membranous inter-ventricular septal defect was noted in subaortic region with ostium secundum type of atrial septal defect, overriding of aorta and biventricular hypertrophy consistent with the diagnosis of Tetralogy of Fallot (TOF). Thickening of atrio-ventricular cusps with severe aortic stenosis and moderate aortic regurgitation was noted. Trivial mitral regurgitation, moderate tricuspid regurgitation and moderated pulmonary arterial hypertension were also present. Multiple vegetations on pulmonary valve and along right and non-coronary sinus were noted suggesting possibility of infective endocarditis. Cardiac CTA was advised for better demonstration of the complex cardiovascular morphology, especially the pulmonary and coronary arterial anatomy as well as detection of major aorto-pulmonary collateral arteries (MAPCAs).

Patient was advised a caffeine-free diet starting from one day before the procedure. Oral beta blocker (Metoprolol-50, repeated after 1 h as HR was not controlled) was given one hour prior to the procedure to reduce heart rate (below 65 bpm) and ectopics. But the patient had occasional ectopics so retrospective ECG-gated cardiac CT angiography was performed on a 64 slice-MDCT scanner (Extended Brilliance Workspace, Version 6.4, Philips Medical System), which worked at 64 mm × 1 mm collimation, with a minimum slice thickness of 0.625 mm operating at 120 kV and 320 mAs. Scanning was performed after the intravenous (iv) administration of a single bolus of 90 ml of non-ionic contrast material (iohexol 350 mg I/ml), with a 30 ml saline chase injected with a MEDRAD power injector (Bayer HealthCare) at the rate of 3 ml/s through an 18-gauge iv catheter placed in left antecubital vein. ECG-gated tube current modulation was applied to decrease the radiation dose to the patient. Appropriate scan timing was determined using bolus-tracking method by placing a region of interest in the ascending aorta. Multiplanar reformations (MPR) and three-dimensional (3D) volume rendered reconstructions in the diastolic phase were used for a detailed morphological assessment.

Cardiac CT confirmed the ECHO features of TOF and well demonstrated the complex anatomy of heart. MAPCAs were absent. An additional and interesting finding was presence of a single large coronary arterial trunk arising from left aortic sinus. Soon after its origin, this

arterial trunk was giving origin to a large left circumflex artery (LCX) running in left atrioventricular groove and terminating into posterior descending artery (PDA). After giving origin to LCX, the arterial trunk continued to run in the anterior inter ventricular sulcus and divided into right coronary artery (RCA) and Left anterior descending artery (LAD). RCA was coursing anterior to RVOT (pre-pulmonic course) to reach the right atrio-ventricular sulcus and giving origin to 3–4 acute marginal branches. LAD was running in the inter-ventricular sulcus to descend down to the notch of cardiac apex (Fig. 1). The patient was referred to cardiovascular thoracic surgery department for surgical correction of TOF. Aortic valve replacement using mechanic valve prosthesis with pulmonary Commissurotomy was performed under general anaesthesia on cardio-pulmonary bypass support. Post-operative period was uneventful and patient was discharged on 10th post-operative day in satisfactory condition. Her breathlessness improved in post op period and she was doing well till recent follow-up done after 6 months of surgery.

Case 2

A 50-year-old man presented with chief complaints of breathlessness while walking for 6 months. There was no history of chest pain. His heart rate and respiratory rates were within normal limits. His ECG and TTE examinations were within normal limits. Coronary CT angiography was advised to rule out coronary artery disease. Oral beta blocker (Metoprolol-50) was given one hour prior to the procedure to reduce heart rate (below 65 bpm). Firstly, a non-contrast CT for coronary artery calcium scoring was performed which was negative. CTA was performed using the same equipment and contrast agent as mentioned in case 1 but with prospective ECG-gating. CTA revealed the presence of single coronary arterial trunk arising from right aortic sinus and supplying the territories of both RCA and LCA. It was traversing the right atrio-ventricular groove, coursing through the crux of the heart and then reaching the left atrio-ventricular groove giving origin to obtuse marginal branches and continuing as left anterior descending artery in anterior inter-ventricular groove (Fig. 2). No evidence of any calcified or soft plaque was noted within the coronary

arteries. The patient was managed conservatively with emphasis on aggressive control of risk factors.

Discussion

SCA is a rare anomaly and is usually detected incidentally while imaging the heart for other reasons. It can be an isolated anomaly or associated with other congenital cardiac anomalies. Our first case was associated with other congenital cardiac anomalies, whereas the second case showed isolated SCA anomaly. When it is associated with anatomically normal heart, it usually divides into normally formed and distributed coronary arterial branches. But cases associated with other congenital cardiac anomalies have atypical branching patterns [2, 3]. A large study on 126,595 individuals revealed that 0.019% had SCA and 40% of them had associated congenital heart diseases like TOF, truncus arteriosus and transposition of great arteries [2]

In another study, performed over 4,445 patients who underwent coronary CT angiography, 12 patients were diagnosed with a SCA with a prevalence of 0.27%. Of the 12 patients with SCA, only one patient had SCA originating from the left coronary sinus, whereas 11 had SCA originating from the right coronary sinus. Dual LAD variant was identified in four patients. None of the patients had associated congenital cardiac anomalies [9].

Most of the patients with SCA are asymptomatic but some can present with chest pain, myocardial infarction, syncope, ventricular tachycardia and even sudden cardiac death [12].

SCA with an inter-arterial (also called malignant) course has been strongly linked with myocardial ischemia and sudden cardiac death, especially among young athletes. The postulated theories include occlusion due to compression between the aorta and pulmonary artery during physical exertion or presence of a more slit-like orifice which is more prone to get occluded [13, 14].

These patients can also present with symptoms of coronary artery disease. It can be related to increased incidence of atherosclerosis among patients with SCA which can be explained by abnormal origin, long travelling distance, anomalous course and compression between the great vessels which may precipitate endothelial injury and eventually lead to atherosclerosis [15].

(See figure on next page.)

Fig. 1 A 19-year-old female with a single left coronary artery associated with Fallot tetralogy: CTA images in **a** axial plane shows thickened pulmonary valve (PV) with prepulmonic course of RCA and dilated ascending aorta (AO) **b** Sagittal plane shows pulmonary stenosis (*) at the level of right ventricular outflow tract (RVOT) and right ventricular hypertrophy (RVH). **c** Axial plane shows thickened and deformed aortic valve (AV) and dilated right atrium (RA). **d** Coronal plane shows subaortic VSD (→) and overriding of aorta. CTA Volume rendered (**e, g, h**) and Maximum Intensity Projection (**f**) images showing a coronary arterial tree with L-II A type of SCA anomaly arising from left Aortic (AO) sinus. Soon after its origin, arterial trunk was giving origin to a large left circumflex artery (LCX) and then continued to run in the anterior inter ventricular sulcus and divided into right coronary artery (RCA) and Left anterior descending artery (LAD). RCA was coursing anterior to RVOT (pre-pulmonic course). PT—pulmonary trunk

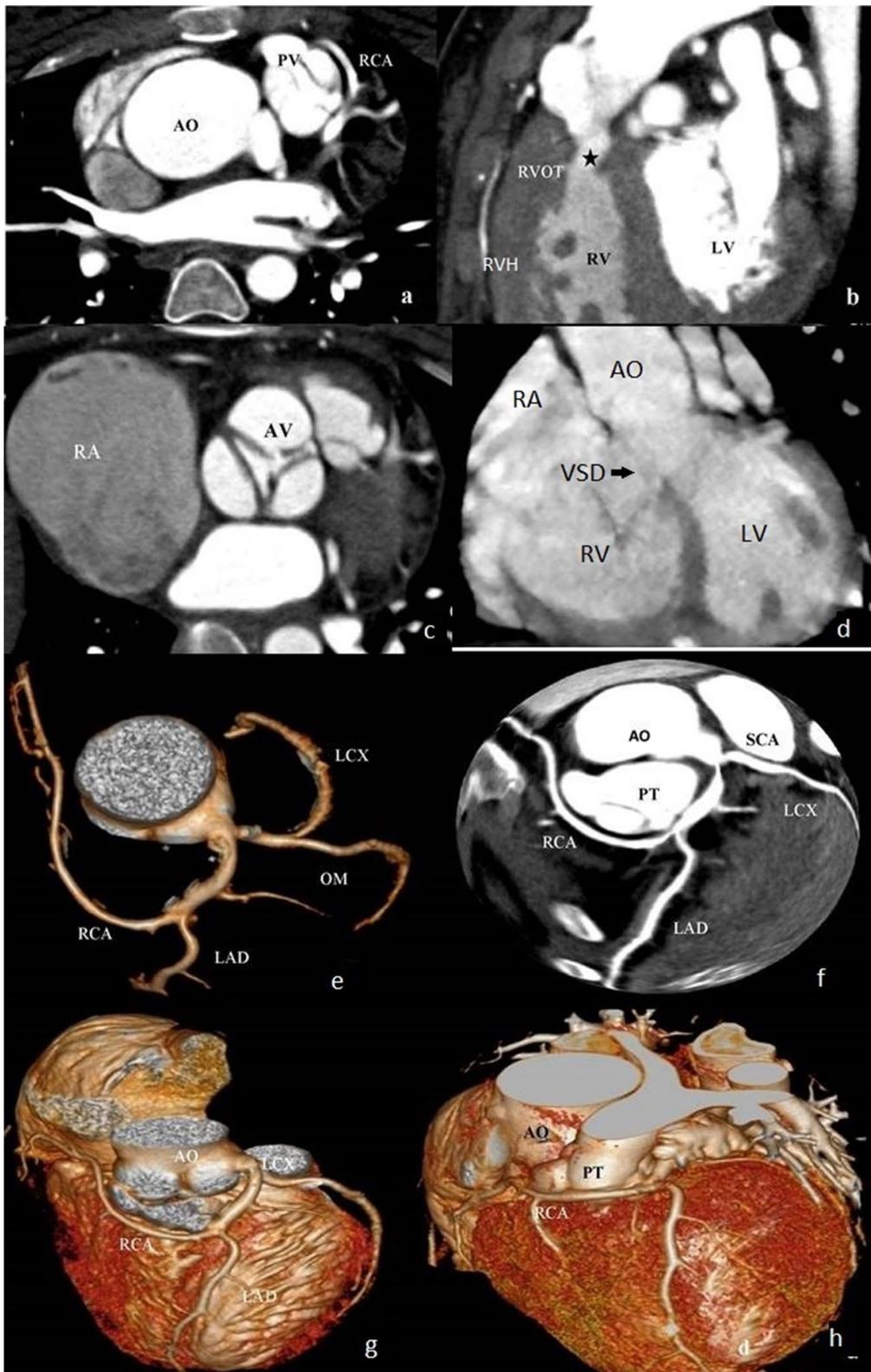


Fig. 1 (See legend on previous page.)

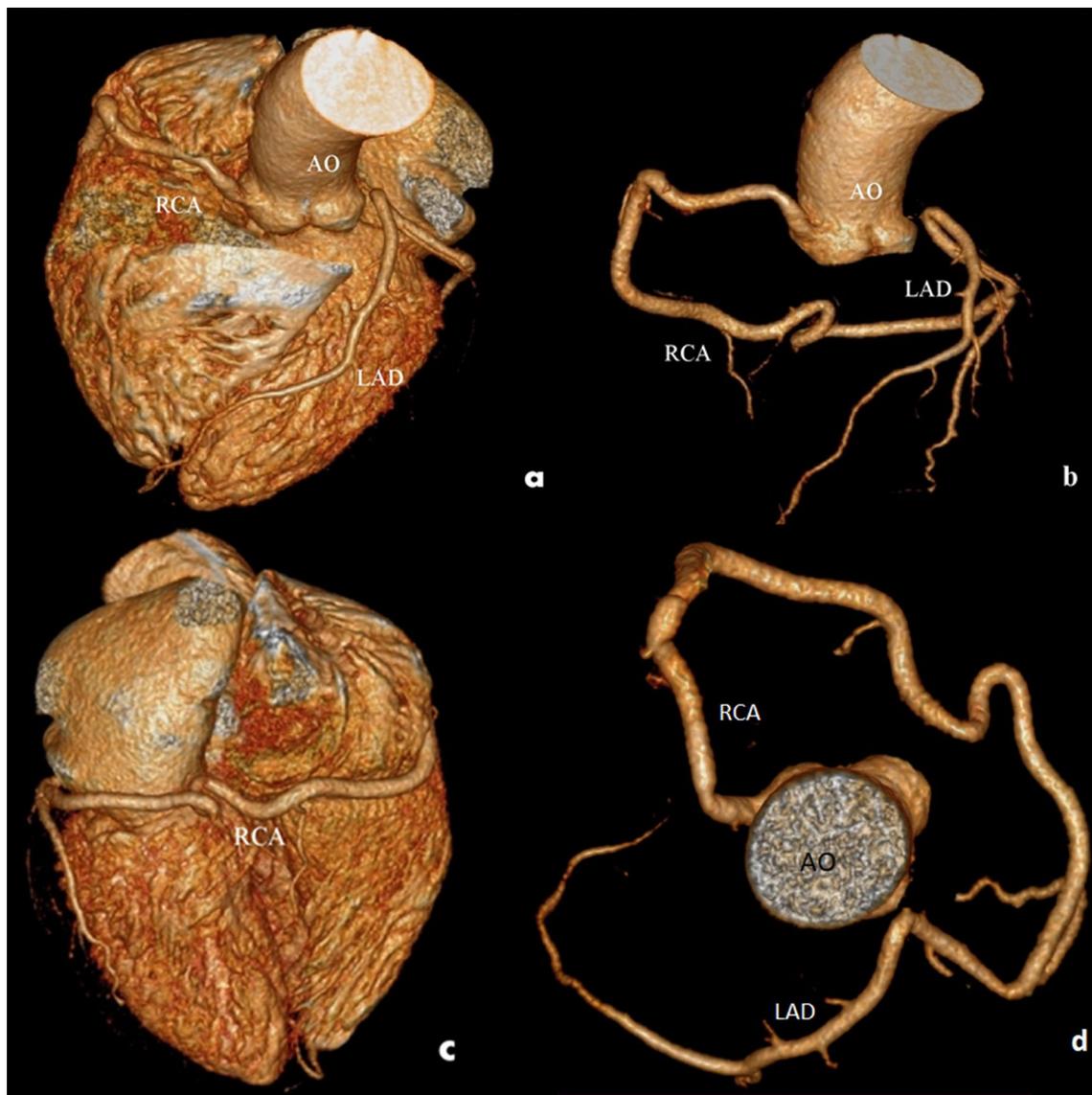


Fig. 2 A 50-year-old man with an isolated anomaly of a single right coronary artery: CTA volume rendered (a–d) images showing coronary arterial tree with isolated R-I type of single right coronary artery (RCA) arising from right Aortic (AO) sinus. It was traversing the right atrio-ventricular groove, coursing through the crux of the heart and then reaching the left atrio-ventricular groove and continuing as left anterior descending artery (LAD)

Previously conventional angiography was considered the gold standard for evaluation of coronary artery anomalies [7]. With advancements in CT technology, Coronary CTA has emerged as the modality of choice for evaluation of coronary arteries. Due to its high spatial resolution and three-dimensional reconstruction abilities, it provides accurate angiographic information about the origin, course, and termination of coronary anomalies non-invasively [16]. Cardiac magnetic resonance imaging (MRI) is an alternative modality to determine coronary artery anomalies; however, due to low spatial

resolution, it is less useful in evaluating the distal coronary system [17].

Having knowledge of the presence of SCA and its course will be helpful in assisting the physician in deciding the treatment strategy and avoiding potential surgical complications. In the majority of asymptomatic individuals without atherosclerotic disease, no invasive intervention is required [12]. Invasive or surgical interventions are recommended for patients with an inter-arterial course of the anomalous artery or symptomatic patients [18]. Surgical procedures include transposition

of coronary arteries to appropriate coronary sinus or coronary artery bypass grafting [19]. None of our patient required surgical intervention for SCA although first case got operated for associated cardiac anomalies and preoperative information about pre-pulmonic course of the coronary artery helped the surgeons in deciding the operative plan.

Conclusions

SCA is a rare anomaly and may be associated with catastrophic life threatening complications. The accurate delineation of the origin and course of the anomalous vessel is of paramount importance when surgical intervention is required as it helps in avoiding potential surgical complications. Management usually involves a multi-disciplinary approach with cardiologists and cardiac surgeons aiming for deciding individual plan based on presentation and anatomy of each case.

Abbreviations

CAA: Coronary artery anomalies; SCA: Single coronary artery; LCA: Left coronary artery; CTA: Computed Tomography Angiography; LCX: Left circumflex artery; RCA: Right coronary artery; LAD: Left anterior descending; TOF: Tetralogy of Fallot; MAPCAs: Major aorto-pulmonary collateral arteries; AO: Aorta; RVH: Right ventricular hypertrophy; 3D: 3-Dimensional; MPR: Multiplanar reformations.

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Author contributions

NS was involved in conceptualization and manuscript writing. YG was involved in image formation. BS and YG were involved in data collection. GR and SR were involved in proof reading of the manuscript. All authors have read and approved the manuscript.

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Availability of data and materials

The datasets used during the current study can be made available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Approval granted by Dr Ram Manohar Lohia Institute of Medical sciences Lucknow Institute ethics committee, IEC no. 37/22. Requirement of consent to participate was waived by the Institute Ethics Committee.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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