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Review Article

Double right coronary artery and its clinical implications

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Abstract Congenital anomalies of the coronary arteries are present in 0.2–1.4% of the general population. These anomalies represent one of the most confusing issues in the field of cardiology and challenges for interventional cardiologists and cardiac surgeons if the anomalies are unrecognised. Double right coronary artery is one of the rarest coronary arteries. Previously, the probability of developing atherosclerotic changes in patients with a double right coronary artery was considered to be equal to that in those without it. In reality, however, a high prevalence of atherosclerotic coronary artery disease was found in patients with a double right coronary artery originating from a single ostium after our comprehensive literature search through the PubMed database. Owing to the fact that double right coronary artery is both a congenital and potentially atherosclerotic coronary artery disease at diagnosis, coronary intervention or cardiac operation is more complicated than previously believed. Individuals with a double right coronary artery may be unaware of its presence until an accidental finding during coronary angiography or cardiac operation and are at risk for unsuspected complications of atherosclerotic coronary artery disease or during cardiac operation. Therefore, it is important to obtain information on the anatomic variants of this congenital coronary anomaly in patients who are undergoing either coronary intervention, aortic root operation or myocardial revascularisation. To our knowledge, this is the first comprehensive article to discuss the anomalies and their clinical implications.

Keywords: Congenital anomalies of the coronary arteries; double right coronary artery; split right coronary artery; coronary artery disease; percutaneous coronary intervention

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CONGENITAL ANOMALIES OF THE CORONARY ARTERIES are present in 0.2–1.4% of the general population.[1] These anomalies represent one of the most confusing issues in the field of cardiology and challenges for interventional cardiologists and cardiac surgeons if the anomalies are unrecognised. Double right coronary artery is one of the rarest coronary anomalies. In a large study, Harikrishnan et al[2] performed 7400 conventional coronary angiographies and reported 34 patients (0.46%) with congenital anomalies of the coronary arteries; a double right coronary artery was detected in only one patient, with an incidence of 0.01%. In the largest series on coronary angiography, which involved 126,595 patients, Yamanaka et al[3] reported a rate of 1.6% for congenital anomalies of the coronary arteries, but no case of a double right coronary artery was mentioned. Kunimasa et al[4] reported that in 2957 consecutive patients undergoing coronary multi-detector-row computed tomography, two (0.07%) presented with a double right coronary artery.

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Double right coronary artery

Double right coronary artery has been described as a right coronary system formed of two distinct

branches, and the two double right coronary arteries have similar diameters.[5] In some patients, a double right coronary artery can originate from a single ostium and split into two branches after a variable short distance from the proximal trunk (Fig 1a), whereas in others it originates from different ostia in the right sinus of Valsalva (Fig 1b). Nevertheless, there are no standard and authoritative definitions of a double right coronary artery until now.[5–8] A comprehensive literature search was carried out through the PubMed database using the keywords "double right coronary artery", "duplicated right coronary artery", "dual right coronary artery", and "split right coronary artery". We also reviewed references from select case reports. So far, a double right coronary artery has been reported 39 times and in 46 patients;[9] among them were 37 men and nine women, with a 4:1 male predominance. The mean age at diagnosis was 54.9 ± 12.1 years. Interestingly, over half (56.5%, 26/46) the cases of this coronary anomaly were reported from Turkey. We divided the patients into two groups based on whether the double right coronary artery originated from a single ostium or from separate ostia in the right sinus of Valsalva.

Double right coronary artery with separate ostia

A double right coronary artery was first reported in the literature in 1987 by Gupta et al.[10] The authors dubbed this anomaly as a "supernumerary right coronary artery". In their study, the two separate vessels arose from the right coronary sinus from two separate ostia adjacent to each other. One of the double right coronary artery vessels had a sinoatrial nodal branch and continued as a posterior descending artery. The other vessel had a conus branch and right ventricular branches, and finally continued as an acute marginal branch.

Overall, there were 18 reported cases of two separate double right coronary arteries of a similar calibre originating from separate ostia in the right sinus of Valsalva (Table 1): 11 cases of "double right coronary artery",[6,11–20] four cases of "duplicated right coronary artery"[21–23] or "dual right coronary artery",[24] two cases of "split right coronary artery",[25] and one case of "supernumerary right coronary artery".[10]

Double right coronary artery with a single ostium

There were 28 reported cases characterising two different right coronary arteries arising from a common

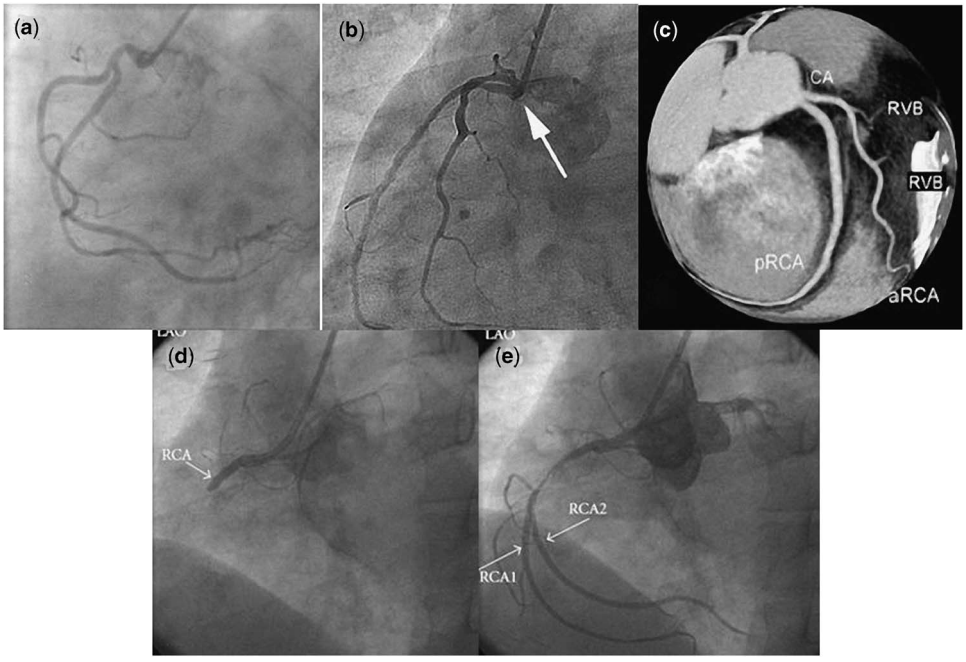

Figure 1.
*(**a**) The right coronary angiography revealed two separate RCAs originating from a single ostium in the right sinus of Valsalva. Both RCAs gave off branches with typical courses and in parallel distribution.[39] (**b**) The posterior anterior view reveals two coronary vessels with almost similar diameters that have adjacent but separated ostia arising from the right coronary sinus.[16] (**c**) The anterior RCA artery gave off two right ventricular branches (acute marginals) ending in the inferoapical area. The posterior RCA had no ventricular branches and gave off two large arteries distally.[18] (**d**) The left anterior oblique view of the totally occluded proximal RCA before percutaneous coronary intervention.[40] (**e**) After predilatation with a sprinter balloon from panel (d), atypical double RCA appeared. (d), atypical double RCA appeared. Reproduced with permission from Selcoki et al,[39] Lemburg et al,[16] Capunay et al,[18] and Akcay et al.[40] CA = catheter angiography; LAO = left anterior oblique; RCA = right coronary artery.*

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Table 1. Collected case reports of double right coronary artery with separate ostia.

						ASVD				CVD		
						RCA		LCA				
Author	Date	No	Sex	Age	Presenting symptom	Ant-	Post-		Treatment	Congenital disease	Acquired disease	Country
Named as supernumerary right coronary												
Gupta[10]	1987	1	M	35	Dyspnoea	N	N	Y	Medically	N	Acute anterior MI	India
Named as double right coronary artery												
Aydogdu[11]	1997	1	M	42	n/a	N	N	n/a	n/a	N	n/a	Turkey
Harikrishnan[12]	2001	1	F	52	Dyspnoea	N	N	N	n/a	N	Severe MS	India
Altunkeser[13]	2001	1	M	34	Chest pain	N	N	N	n/a	N	N	Turkey
Timurkaynak[14]	2002	1	M	54	Chest pain	Y	Y	Y	CABG recommended	N	Acute inferior MI	Turkey
Garg[15]	2002	1	M	52	Dyspnoea	N	N	N	n/a	LM and septal arteries from the RCS	N	India
Lemburg[16]	2007	1	M	46	Chest pain, dyspnoea	N	N	Y	Stent in ramus intermedicus	N	Old antelateral MI	Germany
Karabay[17]	2007	1	M	61	Dyspnoea	N	N	N	n/a	N	N	Turkey
Topaloglu[6]	2007	1	M	48	Chest pain, dyspnoea	N	N	N	Medically	VSD and one RCA arising from the LM trunk	N	Turkey
Capunay[18]	2010	1	M	53	Chest pain	N	N	N	n/a	N	N	Argentina
Astan[19]	2010	1	M	70	Chest pain	N	N	Y	n/a	N	LAD stenosis	Turkey
Chen[20]	2011	1	F	64	Cough	N	N	Y	Medically	Superior RCA–MPA fistula	LCA giant aneurysm	Taiwan
Named as duplicated or dual right coronary artery												
Egred[21]	2005	1	M	46	Chest pain	N	N	N	Medically	N	N	United Kingdom
Karaosmanoglu[22]	2008	1	M	50	Chest pain	N	N	N	Lifestyle change	N	N	Turkey
Ciftci[23]	2009	1	M	31	Chest pain, dyspnoea	N	N	N	Medically	HCM and myocardial bridging	N	Turkey
Named as split right coronary artery												
Huang[24]	2008	1	M	43	Chest pain	N	N	N	Medically	Posterior RCA–RA fistula	N	China
Chen[25]	2010	2	M	69	Chest pain	N	N	N	Medically	N	N	Taiwan
			M	75	Chest pain	N	N	N	Medically	N	N	Taiwan

Ant = anterior; ASVD = atherosclerotic vascular disease; CABG = coronary artery bypass surgery; CVD = cardiovascular disease; F = female; HCM = hypertrophic cardiomyopathy; LAD = left anterior descending; LCA = left coronary artery; LM = left main; M = male; MI = myocardial infarction; MPA = main pulmonary artery; MS = mitral stenosis; N = no; n/a = not available; Post- = posterior; RA = right atrium; RCA = right coronary artery; RCS = right coronary sinus; VSD = ventricular septal defect; Y = Yes

ostium in the right sinus of Valsalva, with a variable short distance from the proximal trunk (Table 2). In 1994, Barthe et al[26] were the first to report two different right coronary arteries arising from a common ostium, coursing down the right atrioventricular groove; they were also the first to use the term "double right coronary artery". Subsequently, this term has become popular and has been adopted by many authors to label two separate double right coronary arteries of a similar calibre originating from either a single ostium or from separate ostia in the right sinus of Valsalva.[12,27] Of the 28 reported cases originating from a single ostium, 23 were described as a "double right coronary artery"[4,26–43] and five as a "split right coronary artery".[44–46]

Imaging studies for double right coronary artery

Correctly diagnosing this rare anomaly has difficulties. It is almost always accidentally detected during traditional coronary angiography. Altun et al[27] reported that the right anterior oblique view during coronary angiography provides the best possibility for differentiating a double right coronary artery from a high take-off of a large right ventricular branch. Several authors have reported the importance of using multi-detector computed tomography for diagnosing this rare congenital anomaly.[4,47,48] Multi-detector computed tomography is a cost-effective, non-invasive, and fast imaging tool that is more accurate for defining the origins and course of anomalous coronary artery compared with conventional coronary angiography[48] (Fig 1c). However, multi-detector computed tomography can be also harmful because it exposes the patient to considerable amounts of radiation and contrast medium.[48] Magnetic resonance coronary angiography is another non-invasive diagnostic tool for assessing proximal coronary anatomy.[49] Magnetic resonance imaging holds the greatest appeal because no radiation and less nephrotoxic contrast agents are used.[37]

Double right coronary artery and atherosclerosis

Overall, there were 46 reported cases of two separate double right coronary arteries with similar calibre arising from the right sinus of Valsalva and originating from either a single ostium or from separate ostia. Previously, the double right coronary artery[15,28,35,36,47,48] or split right coronary artery[46] had been described as a rare benign congenital coronary anomaly that did not warrant special management. Nevertheless, because of the rarity of this anomaly and lack of comprehensive studies, its clinical importance is not yet known. Garg et al[15] reported that a double right coronary artery does not seem to increase the tendency towards atherosclerosis or induce ischaemia. Furthermore, Sato et al[48] suggested that the clinical significance of double right coronary artery might be minimal because the probability of development of atherosclerosis in patients with a double right coronary artery was equal to that in those without it. Nevertheless, chest pain as the initial presenting symptom was present in 69.6% (32/46) of the reported cases. Imaging studies demonstrated significant stenotic lesions in 26.1% (12/46) of anterior right coronary arteries. Surprisingly, the single ostium group had a much higher incidence (39.3%, 11/28) of stenotic lesions than did the separate ostia group (5.6%, 1/18) (odds ratio [OR], 11.0; 95% confidence interval [CI], 1.28–94.89; $p = 0.011$) (Table 3 and Fig 1d and e). This result is consistent with the fact that atherosclerotic lesions occur predominantly at sites of low or oscillatory shear stress patterns such as near bifurcation or curved arteries.[50] In contrast, the straight arterial segment exposed to a physiologic shear stress appeared to be protected from atherosclerosis.[51] In addition, 32.1% (9/28) of the patients in the single ostium group had significant coronary stenosis of the posterior right coronary artery compared with only 5.6% (1/18) in the separate ostium group (OR, 8.1; 95% CI, 1.02–70.33; $p = 0.033$), and 17.9% (5/28) of the proximal main trunks in the single ostium group showed significant coronary stenosis. The incidence of coronary artery disease involving the right coronary arteries in patients with double right coronary artery was 37.0% (17/46). Interestingly, the occurrence of atherosclerotic stenosis of both right coronary arteries was only 5.6% (1/18) in the separate ostia group, whereas it was up to 57.1% (16/28) in the single ostium group, with the difference being highly statistically significant (OR, 22.7; CI, 2.64–194.82; $p < 0.0001$). The overall incidence of coronary artery disease involving the right and left coronary arteries was 54.3% (25/46): 27.8% (5/18) in the separate ostia group and 71.4% (20/28) in the single ostium group, with the difference being highly statistically significant (OR, 6.5; CI, 1.74–24.27; $p = 0.004$). We found no difference as regards the occurrence of coronary artery disease involving the left coronary artery between two groups (OR, 1.4; CI, 0.40–5.24; $p = 0.575$). Of the patients with double right coronary artery, six had complications with acute inferior wall infarction[14,35,36,40,42,44]: five in the single ostium group and one in the separate ostia group. Of the infarcted vessels, four were anterior right coronary arteries and two were posterior right

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Table 2. Collected case reports of double right coronary artery with a single ostium.

						ASVD					CVD		
						RCA			LCA				
Author	Date	No	Sex	Age	Presenting Symptoms	Main trunk	Ant-	Post-		Treatment	Congenital disease	Acquired disease	Country
Named as double right coronary artery													
Barthe[26]	1994	1	F	66	Syncope	N	N	N	N	n/a	N	Severe AS	Spain
Altun[27]	2002	2	M	38	Chest pain	N	N	N	N	n/a	N	N	Turkey
			M	65	Chest pain	N	N	N	Y	n/a	N	N	Turkey
Nair[28]	2005	1	M	45	Chest pain	N	N	N	Y	n/a	N	Acute inferoposterolateral MI	India
Resatoglu[30]	2005	1	M	50	Chest pain, dyspnoea	N	N	Y	Y	CABG and fistula repaired	Superior RCA–RV fistula	N	Turkey
Ozeren[31]	2005	1	M	56	Chest pain, dyspnoea	N	Y	N	N	Medically	N	Sustained VT	Turkey
Erbagci[32]	2006	1	M	50	Chest pain	N	N	N	N	n/a	N	N	Turkey
Sari[33]	2006	1	F	84	Chest pain	Y	Y	Y	Y	CABG recommended	N	N	Turkey
Kunimasa[4]	2007	2	M	57	MDCT follow-up	N	N	N	Y	n/a	N	Stent in LAD	Japan
			M	76	MDCT follow-up	N	N	N	Y	n/a	N	N	Japan
Gulel[34]	2007	1	M	60	Chest pain	N	Y	N	N	Medically	N	Acute anterior MI	Turkey
Tatli[35]	2007	1	M	52	Chest pain	N	Y	N	N	Medically	N	Acute inferior MI	Turkey
Rohit[36]	2008	1	M	53	Chest pain	N	N	Y	Y	PTCA in RCA	N	Acute inferior MI	India
Sari[29]	2008	2	M	50	Chest pain	N	N	Y	Y	n/a	N	Stent in LAD	Turkey
			M	74	Chest pain	N	Y	Y	Y	Stent in anterior RCA	N	Inferior wall ischemia	Turkey
Misuraca[37]	2009	1	F	54	Follow-up	N	Y	N	N	Medically	N	Severe MS	Italy
Sucu[38]	2009	1	F	50	Chest pain	N	N	Y	N	Stent in posterior RCA	N	N	Turkey
Selcoki[39]	2010	2	F	40	Dyspnoea	N	N	N	N	n/a	N	Inferior leads stress test positive	Turkey
			F	66	Chest pain, dyspnoea	N	N	N	N	n/a	N	N	Turkey
Akcay[40]	2010	1	M	40	Chest pain	Y	Y	N	N	Stent in RCA	N	Acute inferior MI	Turkey
Ciftci[41]	2011	1	M	61	Chest pain	N	N	N	N	n/a	LCA arising from a double RCA	N	Turkey
Acet[42]	2012	1	M	61	Chest pain	Y	Y	Y	N	Stent in RCA	N	Acute inferior MI	Turkey
Singh[43]	2012	1	M	70	Syncope	Y	Y	Y	Y	Stent in RCA and LCX	Complete heart block	N	India
Named as split right coronary artery													
Sawaya[44]	2008	1	M	64	Chest pain	N	Y	N	N	Stent in anterior RCA	N	Acute inferior MI	United States of America
Andreou[45]	2010	2	M	n/a	n/a	Y	Y	N	n/a	n/a	N	n/a	Cyprus
			M	n/a	n/a	N	N	N	n/a	n/a	N	n/a	Cyprus
Okmen[46]	2010	2	M	59	Chest pain	N	N	N	N	Medically	N	N	Turkey
			F	50	Chest pain	N	N	Y	N	Medically	N	N	Turkey

Ant = anterior; AS = aortic stenosis; ASVD = atherosclerotic vascular disease; CABG = coronary artery bypass surgery; CVD = cardiovascular disease; F = female; LAD = left anterior descending artery; LCA = left coronary artery; LCX = Left circumflex coronary artery; M = male; MI = myocardial infarction; MS = mitral stenosis; N = No; n/a = not available; Post- = posterior; PTCA = percutaneous transluminal coronary angioplasty; RCA = right coronary artery; RV = right ventricle; VT = ventricular tachycardia; MDCT = Multi-detector computed tomography; Y = Yes

Table 3. Characteristics of atherosclerotic CAD and other associated heart diseases in patients with double right coronary artery.

	n (%)					
Variables	Total (n = 46)	Single ostium (n = 28)	Double ostia (n = 18)	p-value	OR	95% CI
CAD	25 (54.3)	20 (71.4)	5 (27.8)	0.004	6.5	1.74–24.27
RCA*	17 (37.0)	16 (57.1)	1 (5.6)	<0.0001	22.7	2.64–194.82
Ant-	12 (26.1)	11 (39.3)	1 (5.6)	0.011	11.0	1.28–94.89
Post-	10 (21.7)	9 (32.1)	1 (5.6)	0.033	8.1	1.02–70.33
LCA	15 (32.6)	10 (35.7)	5 (27.8)	0.575	1.4	0.40–5.24
CVD	26 (56.5)	17 (60.7)	10 (55.6)	0.729	1.2	0.37–4.10

Ant- = anterior; CAD = coronary artery disease; CVD = cardiovascular disease (i.e. associated with other congenital heart disease and acquired heart disease); LCA = left coronary artery; Post- = posterior; RCA = right coronary artery
*The number of RCA with CAD does not necessarily equal that of Ant- plus Post- because the same patient may have both Ant- and Post-CAD

coronary arteries. The aforementioned findings are different from the speculation proposed by Angelini,[52] who described that coronary atherosclerotic changes seem to affect only the atrioventricular (or basal) right coronary artery branch. In summary, although there are no documented studies demonstrating that people with a double right coronary artery are more susceptible to atherosclerotic changes, the possibility of an association between this condition and the increased susceptibility of developing atherosclerosis and or even acute coronary syndromes, including myocardial infarction, remains to be elucidated, especially in patients having double right coronary artery with a single ostium.

The patients report by Selcoki et al[39] and Garg et al[15] had ST-segment depression in the inferior leads at exercise stress but their coronary arteries were free of atherosclerotic lesions. Thus, in some patients without atherosclerotic stenosis, ischaemia can be a result of anatomical malformation, including the acute take-off angle of the anomalous vessel, with a narrowed slit-like orifice that collapses in a valve-like manner, vasospasm, or myocardial bridging, thereby limiting the blood flow.[31,39]

Issues during percutaneous coronary intervention

In addition to its being a coronary abnormality, we speculate that double right coronary artery originating from a single ostium might be atherosclerotic and can cause acute coronary syndrome. Coronary interventionists should be aware of this anomaly so as to avoid misinterpreting coronary angiograms and potential procedurally induced complications. According to an interesting double right coronary artery imaging report by Misuraca et al,[37] a coronary angiographic view mimics a coronary dissection because the two separate right coronary arteries arise from a single ostium. The two right coronary arteries were split along both the proximal and the mid-segment and united into a single vessel at the distal segment. The incorrect diagnosis of coronary dissection may potentially be arrived at, and the patient may potentially be mismanaged with harmful treatment, such as the application of a stent. Okmen et al[46] emphasised that it can be challenging to catheterise the correct artery without blocking the blood flow to the other artery. For this reason, they recommend making the first injection of the contrast as non-selective as possible. Pulling back the catheter slightly before the first or final injection may also reveal the second right coronary artery originating from the same ostium or adjacent ostium. Thus, it is important for the interventional cardiologist to keep in mind this congenital coronary abnormality, in order to prevent an inaccurate diagnosis and incorrect treatment.[33,37,38]

A total of three patients in the single ostium group had severe atherosclerotic lesions in the double right coronary artery, but percutaneous intervention was difficult because of the highly angulated anatomy of the main trunk of the right coronary system,[33] localisation of the stenotic lesion,[31] or small size of the right coronary artery.[35]

Issues during cardiac surgery

Owing to the fact that the surgeon may improperly manage a pre-operatively undefined abnormal coronary artery during surgery, pre-operative recognition of this congenital anomaly is of paramount importance before using cardioplegia during cardiac operation for aortic valve surgery or an aortic root procedure. All the coronary arteries have to be cannulated separately while delivering cardioplegic solution, or they need to be carefully relocated during aortic root surgeries such as the Jatene operation, modified Bentall procedure, valve-sparing root replacement, or the Ross procedure. In addition, cardiac surgeons should also be careful during coronary revascularisation because both the

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right coronary arteries are usually of similar calibre and size and because their courses may be parallel to each other.[6,32,46]

Study limitation

Conclusions that can be drawn from our studies are limited by a number of factors. First, our database may be flawed by sampling errors as most reports of this congenital abnormality are detected from coronary angiography, and therefore the results may not be generalisable to the general population. Second, most of these patients had coronary angiograms performed because of chest symptoms; as a consequence, the analysis of differential incidences of coronary artery disease is doubtful. Third, men are more likely to undergo cardiac catheterisation compared with women due to the prevalence of coronary artery disease among the younger cohort. Finally, catheterisation patients are a group with an increased prevalence of coronary artery disease compared with the general population. Thus, the selection bias in our study series is inevitable.

Conclusions

Double right coronary artery is one of the rarest coronary anomalies. It can be found in isolation or is occasionally associated with other congenital heart lesions. The mean age at diagnosis is during the 6th decade of life. There is a male predominance of ~4:1. Previously, the probability of developing atherosclerotic changes in patients with a double right coronary artery was considered to be equal to that in those without it. In reality, however, a high prevalence of atherosclerotic coronary disease was found in patients with a double right coronary artery originating from a single ostium. Coronary intervention and cardiac operation are more complicated than previously believed because double right coronary artery is both a congenital and potentially atherosclerotic coronary artery disease at diagnosis. Individuals with a double right coronary artery may be unaware of its presence until an accidental finding during coronary angiography or cardiac operation and are at risk for unsuspected complications of atherosclerotic coronary artery disease or during cardiac operation. Therefore, it is important to obtain information on the anatomic variants of this congenital coronary anomaly in patients who are undergoing either coronary intervention, aortic root operation, or myocardial revascularisation. In addition, continuous surveillance for the atherosclerotic change is recommended for patients with a double right coronary artery with or without intervention. Presently, the debate still persists on the definitive definition of the so-called "double right coronary artery". Therefore, it is time we reach a consensus on the nomenclature of this congenital anomaly.[8]

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