Anomalous origin of the right coronary artery from contralateral side: a series of 17 cases

Sağ koroner arterin karşı taraftan anormal çıkışı: 17 vakalık seri

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Abstract

Objective: Anomalous origin of the right coronary artery (RCA) from left sinus of Valsalva (LSV), coursing between the aorta and pulmonary artery can lead to angina pectoris, acute myocardial infarction or even sudden death in the absence of atherosclerosis. Especially young patients have the risk of serious clinical events, but middle-aged-to elderly patients have variable clinical courses. In this study, we presented angiographic variations, clinical findings and long term follow-up data of RCA originating from left side in middle-aged-to elderly patients

Material and Methods: We reviewed the records of 70,850 patients undergoing coronary angiography between 1999-2005 years. At least two invasive cardiologists examined the angiogram of each patient. If necessary, multislice computerized tomography was used for detailed investigation of the RCA. All patients were followed up for approximately 50 months.

Results: Seventeen patients (0,024%) were found to have anomalous origin of RCA from contralateral side. The anomalous RCA originated within the LSV in 12 (71%) patient, from ascending aorta above the LSV in 5 (29%) patients. The retroaortic course of the initial RCA was seen in 2 (12%) patients while 15 (88%) patients showed interarterial course. No atherosclerotic involvement was seen in 9 (53%) patients. Three patients (18%), one of them had no atherosclerotic lesion, underwent CABG due to anomalous RCA. One patient was

also treated by stent implantation to the anomalous RCA. Three patients died during the follow up period due to noncardiac causes. Other 14 patients have been followed up uneventfully. **Conclusion:** Medical treatment is a plausible choice of treatment in anomalous RCA originating from LSV in middle–aged-to elderly patients in the absence of ischemia on 12-lead resting ECG and/or acute coronary syndrome.

Key words: Angiography, Coronary vessel anomalies, Myocardial ischemia, Sudden death

Özet

Amaç: Sağ koroner arterin sol sinüs Valsalva'dan anormal çıkışı, aort ve pulmoner arter arasında seyretmesi, ateroskleroz olmaksızın göğüs ağrısına, akut miyokard enfarktüsüne ve hatta ani ölüme sebep olabilir. Özellikle genç hastalar ciddi klinik olaylar açsından risk altında olsa da, orta-ileri yaştaki hastalarda da farklı klinik seyirler gözlenebilir. Bu çalışmada, orta-ileri yaştaki hastalarda sol sinüsten Valsalva'dan çıkan sağ koroner arterin anjiyografik varyasyonları, klinik bulguları ve uzun dönem bulguları incelenmiştir.

Gereç ve Yöntemler: 1999-2005 yılları arasında farklı merkezlerde koroner anjiyografi yapılan 70.850 hastanın kayıtları en az iki girişimsel kardiyolog tarafından incelendi. Sağ koroner arterin ayrıntılı değerlendirilmesi gerektiğinde, çok kesitli bilgisayarlı tomografi de kullanıldı. Tüm hastalar ortalama 50 ay takip edildi.

Bulgular: Toplam 17 hastada (%0.024) karşı taraftan çıkan sağ koroner arter tespit edildi. Bu hastaların 12 (%71) sinde sağ koroner arter sol sinüs Valsalva'dan, 5 (%29) inde çıkan aortada sol sinüs Valsalva üzerinden çıkmaktaydı. Sağ koroner arterin başlangıç kısmının retro-aortik seyri 2 (%12) hastada gözlenirken, 15 (%88) inde inter-arteryel seyretmekteydi. Hastaların 9 (%53) unda aterosklerotik tutulum gözlenmedi. 3 (%18) hastaya, bunlardan birisinin aterosklerotik tutulumu yoktu, anormal sağ koronere bağlı koroner by-pass yapıldı. Hastalardan birisine de anormal sağ koroner arter nedeniyle stent takıldı. Takip süresince 3 hasta kalp dışı nedenlere bağlı olarak kaybedildi. Diğer 14 hasta olaysız olarak takip edildi.

Sonuç: Sol sinüs Valsalva kaynaklı sağ koroner arter saptanan orta-ileri yaştaki hastalarda, akut koroner sendrom ya da istirahat EKG'sinde iskemi olmaksızın, tıbbi tedavi uygun bir seçenektir.

Anahtar Kelimeler: Ani Ölüm, Koroner Arter Anomalileri, Miyokardiyal İskemi

Introduction

Anomalous origin of the RCA is a rare congenital anomaly that was first described in 1948 by White and Edwards [1]. When RCA arises anomalously from the LSV, most commonly it courses between the aorta and pulmonary artery or right ventricular outflow tract. There is the risk of myocardial ischemia, angina pectoris and myocardial infarction resulting from this course, as well as ventricular fibrillation and sudden death, even in the absence of coronary atherosclerosis [2]. Firstly, Roberts et al described three cases of sudden death resulting from this anomaly [3]. But we must consider that sudden death is most probably seen less than 30 years-old-patients. It is difficult to manage patients in this anomalous group including its rare prevalence and low event rates. In the present study we describe angiographic variations, relatively long term follow-up data (50±22 months) and clinical findings of anomalous RCA originating from contralateral side in adult patient population referred for coronary angiography.

Materials and Methods

Between 1999 and 2005, 70.850 records of adult patients who had been undergone diagnostic coronary angiography were surveyed, retrospectively. The records of these patients were obtained from these cardiology centers: Medical Center of Sani Konukoglu, University of Harran, Technical University of Black Sea, and University of 100th years and Cardiology Hospital of Hanefi Oksuz (4). The study was approved by our institutional ethics committee and all participants gave informed consent.

Before undergoing coronary angiography, patients were screened by non-invasive tests such as treadmill exercise test with Bruce protocol, exercise tallium-201 myocardial single photon emission computed tomography (exercise 201Tl SPECT), except a few patients who were supposed to have acute coronary syndrome. A positive treadmill exercise test was defined as the existence of >1 mm (0.1 mV) horizontal or down slopping ST segment depression at 80 msec after the J point.

Coronary angiography was performed either by the Judkins femoral method or the Sones brachial method in each center above. Initial attempts at selective engagement of the RCA originating from LSV with left Judkins-curve catheter were not successful for some of the patients. Thereafter, 6F right or left Amplatz I-II catheters (Medtronic AV; Galway, Ireland) were introduced successfully. The course of anomalous coronary artery was determined by the help of different coronary angiographic views (Left anterior oblique (LAO), right anterior oblique (RAO) and lateral views). The records of each patient were examined by at least 2 experienced angiographers. In one of the patients (case 6), multislice computerized tomography (MSCT) was performed due to failure of selective catheterization of the anomalous RCA.

The patients were called every 6 months for about 50 months. Their follow-up data were supplied by a questionnaire obtained on the telephone conversation.

Results

Patients and clinical presentations

Among 70.850 patients, anomalous RCA originating from contralateral side was diagnosed in 17 patients (a prevalence of 0,024%). Clinical and angiographic properties of 17 patients were summarized in Table 1. The mean age of 17 patients was 58±8 years (42-74). Ten patients were male (59%), with a mean age of 59±9 years. Seven patients (42%) were female, with a mean age of 56±8 years.

Nine of the 17 patients had no atherosclerotic coronary artery disease (52%). Five of the 9 patients had typical stable angina pectoris and 3 of them had symptom positive exercise tests, 2 of them had positive exercise tests. Two of the 9 patients (cases 6 and 15) were presented with suspicious acute coronary syndrome diagnosis. Coronary angiography was carried out because of dynamic ECG changes. Although Cases 11 and 12 had atypical chest pain, they underwent coronary angiography because of their symptom positive stress tests.

Eight patients (47%) were associated with atherosclerotic coronary artery disease (defined as more than 50% luminal stenosis of 1 or more major epicardial coronary arteries). Five of the 8 patients had typical stable angina pectoris and 3 of them had positive exercise tests, 2 of them had

positive SPECT. Three of the 8 patients were presented as acute coronary syndrome.

Coronary angiographic characteristics:

In 12 patients (71%) (Figures 1, 2), the anomalous RCA originated from LSV. In other 5 patients (Figures 3), the anomalous RCA originated from ascending aorta above the LSV (29%). The distance between the coronary origin and the left sinotubular junction demonstrated a range of ~0.5 to 5 cm. One of these cases had acute coronary syndrome. His coronary angiography showed significant atherosclerosis and coronary artery aneurysm in mid-RCA (Figure 4) and significant atherosclerosis in mid-circumflex artery. The most common involved coronary atherosclerotic lesions were in the left anterior descending artery (LAD) and circumflex artery (CX) (41%). Atherosclerotic lesions were encountered in the anomalous RCA in 3 of patients (18%). Right coronary dominance was seen in 14 patients (82%) (Table 1).

Initial course of the anomalous RCA was retroaortic in two patients (12%) while others course between the aorta and pulmonary trunk (88%) (Figure 5). There was no patient with an anomalous RCA coursing ahead of pulmonary trunk. We needed a confirmation by MSCT imaging in only one case that had experienced acute coronary syndrome twice. Selective cannulation of the RCA in the case couldn't be performed in two separate cardiology center (Figure 6). The semi-selective imaging of the anomalous RCA in that case was not sufficient for management of the patient. The diagnosis of the interarterial courses in this study was confirmed surgically in five cases (Table 1).

Treatment and Survival:

Six patients underwent surgical treatment (35%). In 3 patients, CABG was performed to anomalous RCA (18%). One of these patients had no atherosclerotic lesion (Figure 6). A stent was successfully implanted to an anomalous RCA originating from tubular aorta (Figure 4). A medical treatment regimen including beta-blockers, nitrates, calcium-channel blockers, antiplatelets or antiarrhythmic drugs was given to 10 patients for whom revascularization therapy was not considered (59%). Beta-blockers, statins, antiplatelet agents were also given to the 6 patients undergoing CABG and one patient undergoing percutaneous coronary intervention (41%). The symptoms of the patients were improved following medical or revascularization therapy.

The follow-up period ranged from 26 months to 88 months (50±22 months) for the patients who had anomalous RCA originating from LSV. Three cases died during the follow up period due to noncardiac causes (chronic renal failure, cerebrovascular accident, colon carcinoma). There were no deaths directly attributable to anomalous origin of the coronary artery. Surviving 14 patients are taking medical treatments regularly and they are free of symptoms.

Discussion

Coronary artery anomalies are not seen frequently during routine cardiac catheterization. The incidence of anomalous coronary artery is reported to range from 0.2% to 1% of patients undergoing coronary angiography and 0.3% of autopsies (4-6). Angiographic prevalences for anomalous coronary origin from the contralateral sinus (or side) are about 0.04% to 0.33% and anomalous RCA originating from LSV is more common (~73%) than the anomalous LAD originating from the right sinus of Valsalva (RSV)

(Table 2). However the incidence of this anomaly may vary in different populations. An analysis of autopsy reports for the anomalous RCA indicated a prevalence of 0.026% [7]. We found a prevalence of 0.024% while Wilkins et al report a prevalence of 0,28% [8] (Table 2).

Origin of the right coronary ostium above the sinotubular junction of the RSV is considered as a minor variation occurring in about 8% of adult hearts [9]. However, origin of the right coronary ostium from above the sinotubular junction of the LSV is exceedingly rare. Clinical and angiographic properties of patients with high take-off of RCA from the tubular aorta have not been stressed in detail in the literature. There have been a few cases with this anomaly in the literature where sufficient information was provided [10-12]. In this study we presented 5 patients with the anomalous RCA originated from ascending aorta above the LSV (a prevalence of 0,007%) (Figure 3, Table 1). Anomalous origin of the RCA from the contralateral side may be classified further based on the initial course of the anomalous artery. In the majority of cases (67-100%), the anomalous vessel courses between aorta and pulmonary trunk, with the remainder usually coursing posterior to the aorta [5,13]. The RCA in anomalous origin can also course anterior to the pulmonary trunk. Clinically, it is important to determine whether the proximal portion of the abnormal coronary artery is in front of the pulmonary trunk, behind the aorta, or between the two. This distinction has prognostic value. Depending on the coursing anomalous RCA, patients with overt ischemia caused by interarterial coursing often undergo the coronary artery surgery [14].

The coronary angiography is a sufficient method of evaluation in most of the patients with coronary artery anomalies. However, selective coronary angiography cannot always provide the required information adequately. Therefore noninvasive imaging techniques such as MSCT, magnetic resonance imaging, and electron beam tomography are the ways of providing a good anatomical view of the coronary tree (Figure 6).

Most angiographically identified coronary artery anomalies are benign clinically. In contrast, some coronary anomalies are well described as a cause of cardiovascular morbidity and mortality [15]. It is well established that an anomalous origin of the RCA can lead to angina pectoris, myocardial infarction, or sudden death in the absence of atherosclerosis [16]. It accounts for approximately 42% of all potentially malignant anomalies of the coronary arteries5. It should be considered that sudden death is most probably seen less than 30 years-old-patients. The diagnosis and the treatment of this anomaly for the patients under 30 years old must be carried out in detail.

Several theories have been proposed to explain the mechanism of ischemia in patients with anomalous origin of the left coronary artery from the RCA or the right sinus [15]. Similar mechanisms are considered to be related to ischemic events seen in the anomalous RCA cases: (a) compression of the RCA by aorta and the pulmonary trunk, (b) Slit-like orifice produced by acute-angle take-off of the RCA originating from the LSV or the tubular aorta, (c) myocardial squeezing, (d) vasospasm, and (e) small artery. Recently, it has been suggested that the proximal portion of the interarterial RCA might be more prone to spasm [17]. It is also postulated that the severity of the ischemia may be related to how far the ectopic origin is displaced toward the left and

posteriorly, as well as the height of origin above the LSV [11,13]. Several episodes of brief ischemia are probably the cause of substrate for the catastrophic event [18].

There still is not a consensus on the management of anomalous coronary arteries with origin from the contralateral aortic sinus. It is not clear that surgical intervention currently improves long-term outcomes. On the other hand, long-term data about medical management are again lacking. This lack of data is more evident for the patients with the non-interarterial coursing. Without this crucial clinical data, it is undoubtedly difficult to set guidelines/recommendations for surgical correction of this condition. In their recent review, Mirchandani S and Phoon CKL have assessed that current medical opinion is heavily skewed by surgical management of all symptomatic cases, and of many, if not all, asymptomatic pediatric cases with the interarterial coursing [19]. In conclusion, they suggested surgery in patients with anomalous coronary artery with an origin from the contralateral sinus and interarterial coursing who are younger than 30 years old with evidence of ischemia or ventricular arrhythmias, including worrisome symptoms such as syncope. Management of other groups remains controversial. An editorial by Pelliccia recommends that in older asymptomatic patients without inducible symptoms, an incidental finding of wrong sinus origin of a coronary artery probably does not require surgery, because the risk of sudden death occurs at a young age (<30 years) [20]. According to Mirchandani and Phoon [19], all patients in present study were in the controversial groups. This situation is probably true for the majority of the RCA anomalies seen in adults patients >30 years. In Japan, the anomalous RCA has been usually treated medically. Kaku et al. described 44 patients who were treated medically with an anomalous RCA originating from LSV that coursed between the aorta and pulmonary artery or right ventricular outflow tract [21]. In these patients, there were no deaths during the follow up period (mean 5.6±4,2 years). However in the Western world, when an anomalous RCA with interarterial coursing causes angina pectoris in the presence of ischemia and in the absence of atherosclerosis, surgery is more frequently recommended [22]. In the present study, we preferred medical treatment in the absence of associated severe coronary artery disease and/or overt ischemia caused by interarterial coursing.

Conclusion

Coronary artery anomalies are encountered more often on the daily practice of high-volume cardiac centers. Cardiac surgeons and interventional cardiologists should be aware of these anomalies because recognition is mandatory in order to prescribe appropriate medical or surgical therapy. However, there is no sufficient data to optimally manage this specific family of coronary artery anomalies. According to the results of our long-term follow-up data (50±22 months) medical treatment may be adequate in anomalous RCA originating from LSV in middle–aged-to elderly patients in the absence of ischemia restricting functional capacity and/or acute coronary syndrome.

But we still need more data about natural history, clinical findings and angiographic properties of this rarely seen coronary anomaly.

Conflict of interest: None declared

Table 1: Clinical and angiographic properties of the patients with anomalous right coronary artery (AA: Ascending aorta, A/P: Aorta/pulmonary artery, ACP: Atypical chest pain, ACS: Acute coronary syndrome, CABG: Coronary artery bypass grafting, CX: Circumflex artery, LAD: Left anterior descending artery, LMCA: Left main coronary artery, LSV: Left sinus of Valsalva, RCA: Right coronary artery, SAP: Stable angina pectoris, SPECT: Single photon emission computed tomography)

									Atherosclerosis			
Cases	Sex	Age	Symptom	Rest ECG	Ischemia on stress test/SPECT	Origin of RCA	Initial course	RCA	Other coronaries	Dominant coronary	Treatment	Follow up (50±22)
1	M	67	SAP	ST-T wave changes	Treadmill (+)	From LSV	Between A/P	-	LAD, CX	CX	Medical	28
2	F	55	SAP	ST-T wave changes	Treadmill (+)	From LSV	Between A/P	-	-	RCA	Medical	88
3	M	54	ACS	ST-T wave changes	-	From LSV	Between A/P	+	LAD, CX	CX (Thin RCA)	CABG	76
4	M	52	SAP	Diffuse T wave (-)	Treadmill (+)	From LSV	Between A/P	-	LAD, CX	Co-dominance	CABG	76
5	M	58	SAP	ST-T wave changes	SPECT(+)	From LSV	Between A/P	+	LMCA	RCA	CABG	38
6	F	64	ACS	ST-T wave changes	-	From LSV	Between A/P	-	-	RCA	CABG	76
7	M	47	SAP	Inferolateral T changes	Symptom(+)Treadmill	From LSV	Retroaortic	-	-	RCA	Medical	44
8	M	74	ACS	ST-T wave changes	-	From LSV	Between A/P	-	LMCA, LAD, CX	RCA	CABG	37
9	F	66	SAP	-	Symptom(+)Treadmill	From LSV	Retroaortic	-	-	RCA	Medical	36
10	F	55	SAP	ST-T wave changes	SPECT(+)	From LSV	Between A/P	-	LAD, CX	RCA (thin LAD)	Medical	60
11	M	62	ACP	ST-T wave changes	Symptom(+)Treadmill	From LSV	Between A/P	-	-	RCA	Medical	28
12	M	50	ACP	-	Symptom(+)Treadmill	From LSV	Between A/P	-	-	RCA	Medical	26
13	F	42	SAP	-	Symptom(+)Treadmill	From AA above LSV (3 cm)	Between A/P	-	-	RCA	Medical	30
14	F	51	SAP	ST-T wave changes	Treadmill (+)	From AA above LSV (3 cm)	Between A/P	-	-	RCA	Medical	30
15	F	60	ACS	ST-T wave changes	-	From AA above LSV (0,5cm)	Between A/P	-	-	RCA	Medical	77
16	M	66	SAP	ST-T wave changes	Treadmill (+)	From AA above LSV (1 cm)	Between A/P	-	CX, LAD	RCA	CABG	76
17	M	62	ACS	ST-T wave changes	SPECT(+)	From AA above LSV (5 cm)	Between A/P	+	LAD, CX	RCA	Stenting to RCA and CX	28

Table 2 : Prevalences of contralateral origin of coronary arteries, with or without interarterial coursing, in angiographic studies in the literature (LAD: Left anterior descending artery, LCA: Left coronary artery, LMCA: Left main coronary artery, RCA: Right coronary artery)

Study	Sample (n)	RCA % (n)	LCA (LMCA/LAD) % (n)	RCA+LCA % (n)
Yamanaka and Hobbs,1990 (5)	126 595	0,107 (136)	0,047 (22/38)	0,155 (196)
Tuncer et al, 2006 (4)	70 850	0,024 (17)	0,020 (2/12)	0,044 (31)
Gol et al, 2002 (6)	58 023	0,081 (47)	0,040 (17/6)	0,120 (70)
Kaku et al, 1996 (21)	17 731	0,248 (44)	0,011 (1/1)	0,259 (46)
Villa et al, 2001 (23)	13 500	0,111 (15)	0,030 (2/2)	0,141 (19)
Wilkins et al, 1988 (8)	10 661	0,281 (30)	0,047 (3/2)	0,328 (35)
Total	297 360	0,097 (289)	0,036 (47/61)	0,133 (397)

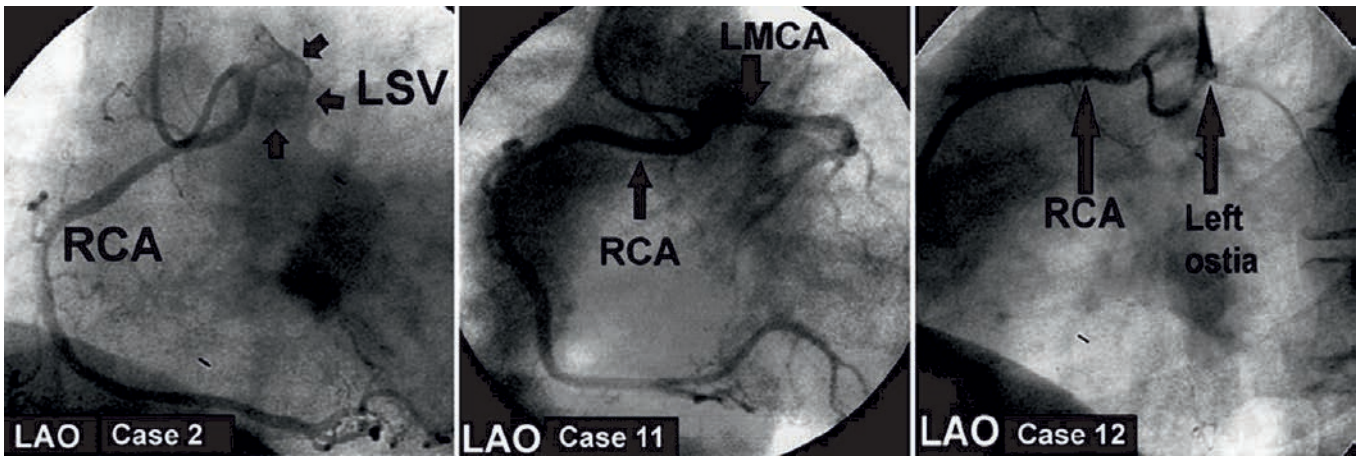

Figure 1: Examples of the anomalous right coronary artery from the left sinus Valsalva (LAO: Left anterior oblique, LMCA: Left main coronary artery, RCA: Right coronary artery).

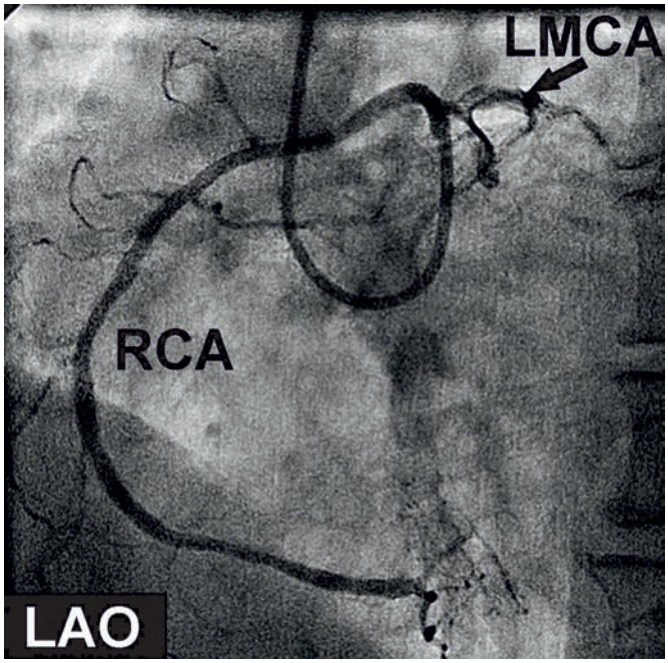

Figure 2: The anomalous right coronary artery arises from the left sinus with acute angulation (LAO: Left anterior oblique, LMCA: Left main coronary artery, RCA: Right coronary artery).

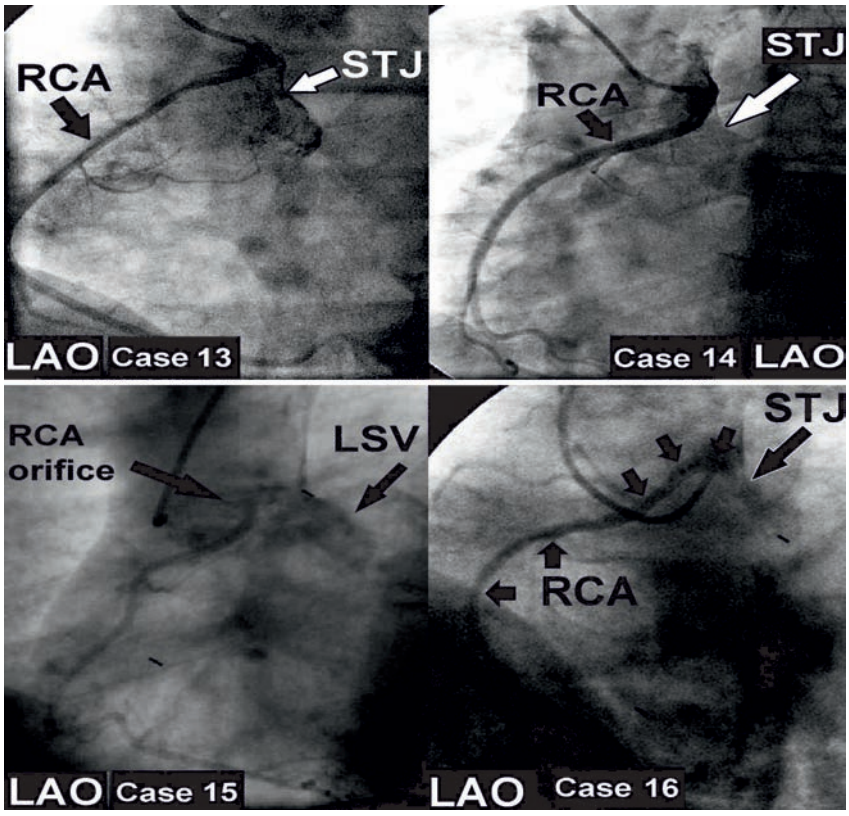

Figure 3: Angiographic variations of the high take-off of the right coronary artery are seen in cases. Acute angulation of the right coronary artery is angiographically seen in cases 13 and 16 only (LAO: Left anterior oblique, LSV: Left sinus of Valsalva, RCA: Right coronary artery, STJ: Sino-tubular junction).

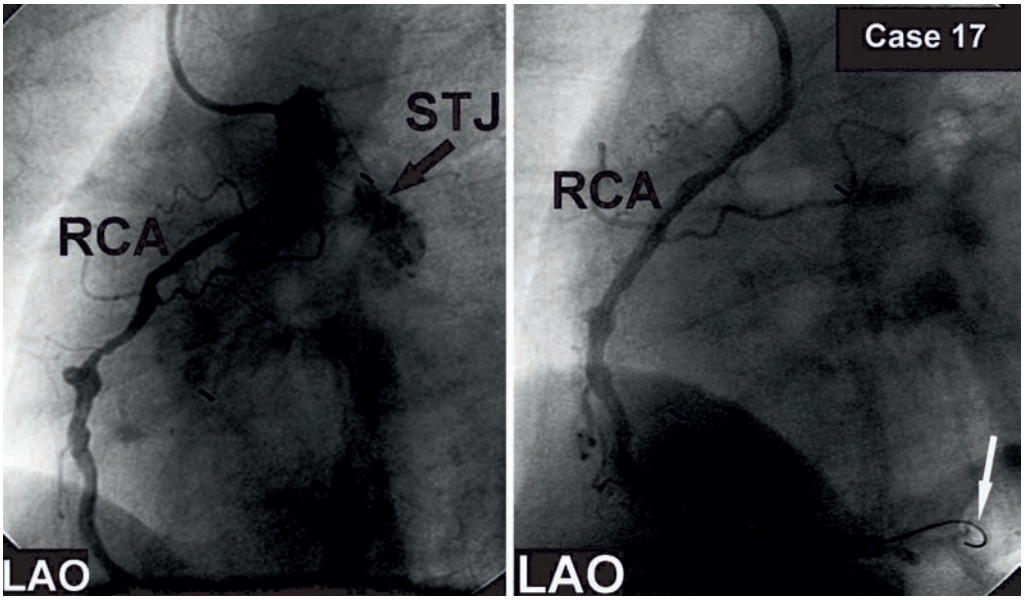

Figure 4: The high takeoff of the right coronary artery is seen. The right coronary artery shows significant atherosclerotic involvement and coronary artery aneurysm. Percutaneous coronary intervention with stenting was successfully performed in this patient (LAO: Left anterior oblique, RCA: Right coronary artery, STJ: Sino-tubular junction).

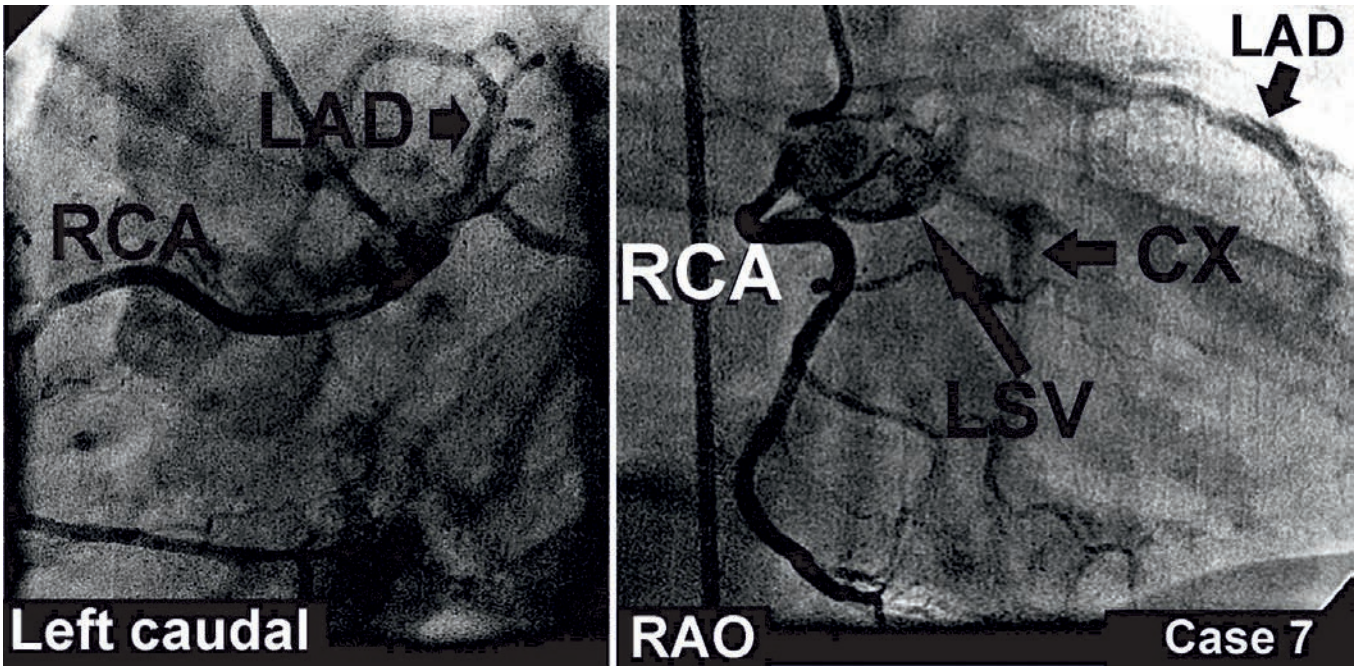

Figure 5: It is seen the retroaortic course of the anomalous right coronary artery originating the left sinus (CX: Circumflex coronary artery, LAD: Left anterior descending artery, LMCA: Left main coronary artery, LSV: Left sinus of Valsalva, RAO: Right anterior oblique, RCA: Right coronary artery).

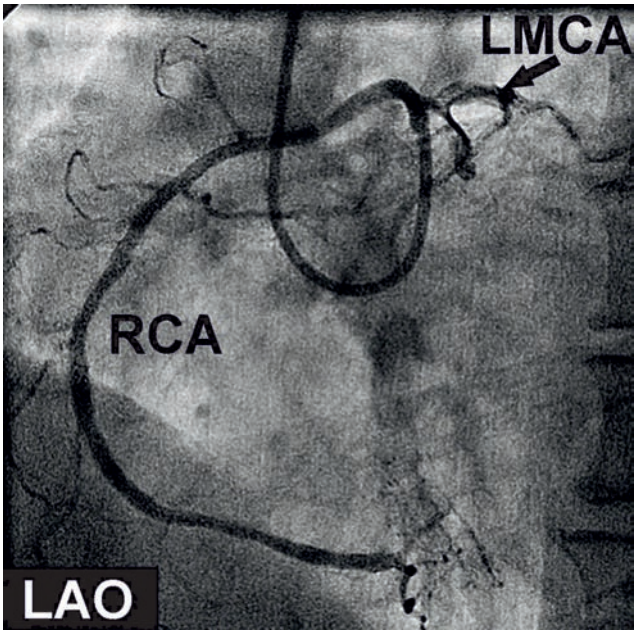

Figure 6. Figures A and B show semi-selectively opacification of the right coronary artery with small coronary orifice and very acute angulation. Figures C-F show the significant compression of the right coronary artery between the great arteries in different slices of the multislice computed tomography (Ao: Aorta, CX: Circumflex coronary artery, LAD: Left anterior descending artery, LAO: Left anterior oblique, LMCA: Left main coronary artery, MPA: Main pulmonary artery, RCA: Right coronary artery, RVOT: Right ventricular outflow tract).

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