

# Diagnosis and Surgical Treatment of the Anomalous Origin of the Coronary Arteries

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## SUMMARY

### Background

Anomalous origin of the coronary arteries is an uncommon congenital heart disease, yet it is important as a potentially preventable cause of sudden death. Treatment of this condition, in particular surgery (or other revascularization procedures), is controversial; however, the development of non-invasive diagnostic techniques allows to make the proper diagnosis more frequently. For this reason, the number of patients diagnosed with this defect is increasing.

### Objective

To analyze a population of patients with anomalous origin of the coronary arteries through the evaluation of diagnostic methods and surgical treatment.

### Material and Methods

Patients undergoing surgery between 2004 and 2010 were retrospectively evaluated. We analyzed the clinical features, symptoms, complementary tests, indication of surgery and techniques used.

### Results

A total of 23 patients (17 men and 6 women) between 18 and 32 years were evaluated due to exertional symptoms: angina (n=12; 52.2%); chest pain (n=4; 17.4%); syncope (n=4; 17.4%), and dyspnea (n=3; 13%). All electrocardiograms were normal, while exercise stress test had positive results in 10 cases.

All patients underwent echocardiographic evaluation; the anomalous origin was detected in 16 patients (69.5%) and the proximal course was identified in 12 (52.2%). The diagnosis was made or confirmed by computed tomography angiography in the 23 patients; the method identified an interarterial course arising from the contralateral sinus.

Surgical techniques were reimplantation of the coronary artery in 7 cases, coronary artery bypass grafting in 3 and unroofing the coronary sinus in 13 cases. No deaths were reported.

### Conclusions

The defect is more common in the origin of left coronary artery. The diagnostic value of electrocardiogram was low.

The diagnosis was made by computed tomography angiography which identified the proximal course.

Unroofing was the surgical technique most commonly used.

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### Key words

> Cardiovascular Anomalies - Tomography - Cardiac Surgery

### Abbreviations

>	MSCT      Multislice computed tomography	AOCA      Anomalous origin of the coronary
	CPB      Cardiopulmonary bypass	arteries

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## BACKGROUND

The importance of the anomalous origin of the coronary arteries (AOCA) has evolved during the last decades from being a simple scientific curiosity to be considered a potentially correctable cause of ischemia and sudden death, particularly in young populations. (1-3)

Treatment of this condition, in particular surgery (or other revascularization procedures), is controversial; however, the development of non-invasive diagnostic techniques, especially echocardiography and multislice computed tomography (MSCT) angiography, allows making the proper diagnosis more frequently. For this reason, the number of patients diagnosed with this defect is increasing. (4, 5)

We retrospectively studied a recent series of patients with AOCA undergoing surgical revascularization in order to analyze the diagnostic tools, the surgical techniques used and the outcomes.

## MATERIAL AND METHODS

Patients with AOCA consecutively undergoing surgical treatment in two university institutions between January 2004 and January 2008 were retrospectively analyzed. The condition was identified by complementary studies.

The following data were evaluated: demographics, clinical history, preoperative assessment, associated comorbidities, and results of complementary tests, surgical data and postoperative outcomes.

The anomalous origin of a coronary artery was defined as the presence of: a coronary ostium originating from the contralateral sinus of Valsalva (the left coronary artery emerging from the right sinus, the right coronary artery originating from the left sinus) or from the non-coronary sinus; a single coronary ostium or a coronary artery originating from the main pulmonary artery or its branches.

The medical records were revised with special consideration to the presence of symptoms and studies performed, and with particular interest about how the diagnosis of AOCA was made. Two independent observers analyzed the clinical data and complementary tests. The therapeutic decision was evaluated based on the results of the tests, patients' characteristics, and on the criteria the different institutions used to define if the coronary anomalies were hemodynamically significant, which included an interarterial course (between the pulmonary artery and the aorta) or an intramural course (a segment of the coronary artery courses in the aortic wall), and the coronary artery originating from the pulmonary artery.

Exclusion criteria: patients < 18 years and those with AOCA associated with other causes (cardiomyopathies, valvular heart diseases, associated coronary artery disease) that required surgical treatment were excluded.

## Surgical techniques

The patients were fasted for more than 4 hours with no ingestion and xanthine medication 12 hours before the study. About 60.5% of the patients who were taking beta-blockers interrupted the medication.

## Basal two-dimensional echocardiogram

Three types of procedures were used:

- Traditional coronary artery bypass graft surgery with or without cardiopulmonary bypass (CPB).
- Unroofing procedure in which the aorta was opened in a transverse fashion approximately 2 cm above the

sinotubular junction. The aortotomy was then extended parallel to the annulus of the aortic valve. At this point, the coronary ostium was identified. When the anomalous vessel was located distant from the aortic commissure, the vessel was unroofed along the common wall with the aorta. The circumference of the tract was then reapproximated and sutures were placed in an interrupted fashion (traditional unroofing technique). When the commissure was in close proximity to the coronary origin, a small coronary probe was passed from the ostium through the anomalous course and a neo-coronary ostium was created in the correct sinus (modified unroofing procedure, Figure 1 A-C).

- Coronary reimplantation: direct coronary ostium reimplantation creating a neo-coronary ostium in the correct coronary sinus; the anomalous orifice was closed to prevent competitive flow. This procedure was used in patients with a coronary artery originating from the pulmonary artery.

All the procedures were performed under general anesthesia and median sternotomy, using CPB, aortic and bicaval cannulation and antegrade and retrograde cardioplegia, except for two patients who underwent bypass grafting without CPB. All patients recovered in the cardiovascular postoperative unit.

## Statistical Analysis

The chi square test or Fisher's exact test were used to compare categorical variables, and the t Student's test was used for continuous variables. The relation between discrete variables is expressed as odds ratio (OR) with its corresponding 95% confidence interval (CI). Quantitative variables are expressed as means  $\pm$  standard deviation. A p value < 0.05 was considered statistically significant.

## RESULTS

A total of 4711 patients underwent surgery during the period evaluated, 23 due to AOCA. Five additional

**Fig. 1.** Modified unroofing technique. **A.** A coronary probe is passed through the slit like ostium. **B.** A round neo-ostium is created. **C.** The procedure is finished.



patients presented AOCA associated with other conditions: mitral valve disease in 4 cases and aortic valve disease in 1. Surgical indication was due to these conditions.

The 23 patients intervened due to AOCA were 17 men and 6 women; age ranged from 18 to 32 years (mean  $24 \pm 6$ ). One patient (4.3%) had a history of hypertension; another was diabetic and 5 (21.8%) were current smokers (Table 1).

All patients had been referred due to exertion symptoms: angina ( $n = 12$ ; 52.2%); atypical chest pain ( $n = 4$ ; 17.4%); syncope ( $n = 4$ ; 17.4%), and dyspnea ( $n = 3$ ; 13%). None of the patients had a history of myocardial infarction and all rest electrocardiograms were normal.

All the 23 patients underwent a maximal exercise stress test, with positive results in 10 (43.5%) cases: 8 patients presented ST-segment depression and 2 had non-sustained ventricular tachycardia.

Transthoracic and transesophageal echocardiography [Phillips IE33, Phillips Ultrasound, USA] was performed to all patients; the anomalous origin was detected in 16 patients (69.5%) and the proximal course was identified in 12 (52.2%).

A MSCT angiography was performed in the 23 patients using a 16-row (in 7 patients) or a 64-row (in 18 patients) scanner (Phillips Medical Systems, Best, the Netherlands). Images were digitized and evaluated in a work station (MXView, Phillips Medical System). This study confirmed the diagnoses of AOCA: the circumflex coronary artery originating from the pulmonary artery in 2 patients (8.7%), the left coronary artery emerging from the right sinus of Valsalva in 16 patients (69.5%) and the right coronary artery originating from the left sinus of Valsalva in 5 patients (Figure 2). The study also allowed the identification of an intraarterial or intramural course in 21 cases arising from the contralateral sinus (Figure 3).

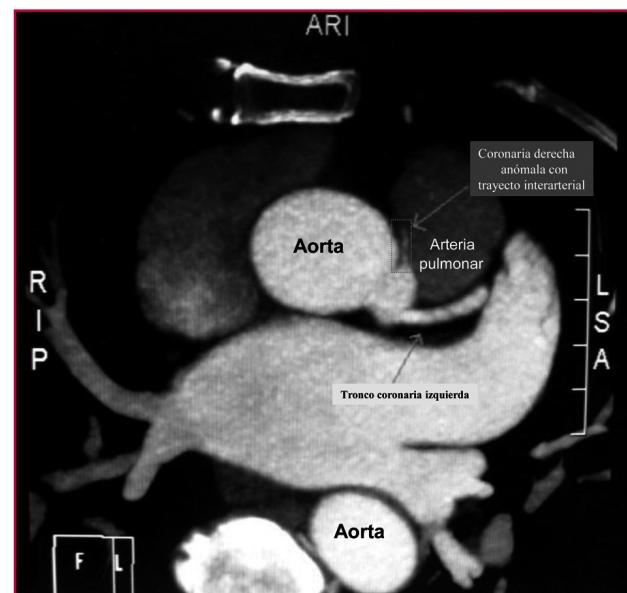
A conventional coronary angiography was performed in 4 patients, confirming the absence of other coronary lesions.

### Surgical procedure

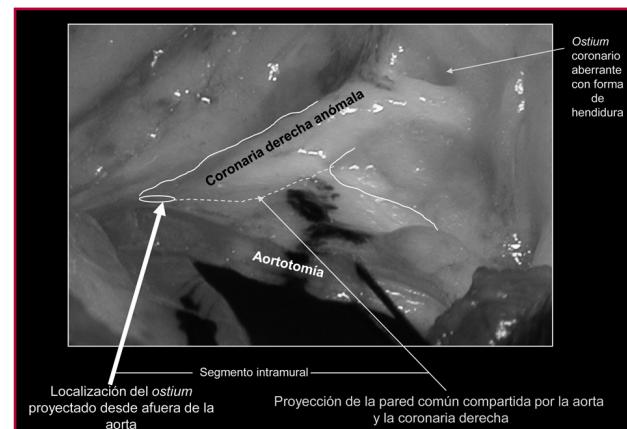
The two patients with left circumflex coronary artery originating from the pulmonary artery underwent coronary ostium reimplantation from the pulmonary artery to the aortic root, ligating the incorrect ostium with a pericardial patch. Two left mammary artery grafts to the left anterior descending coronary artery and two saphenous vein grafts to the left circumflex coronary artery were performed in those cases with anomalous origin of the left coronary artery from the contralateral sinus; CPB was used in one of the procedures. The anomalous coronary ostia were ligated to prevent competitive flow.

In 4 patients the artery was reimplanted in the correct sinus creating a neo-coronary ostium. The traditional unroofing technique was used in

**Fig. 2.** Intraarterial course of the right coronary artery originating from the left sinus of Valsalva.



**Fig. 3.** Projection of the intramural segment of the aberrant right coronary artery (as seen inside the aorta).



**Table 1.** Basal characteristics of the population ( $n = 23$ )

Variables	
Age, years	$24 \pm 6.2$
Male gender, n (%)	17 (73.9)
Previous symptoms, n (%)	23 (100)
Hypertension, n (%)	1 (4.3)
Diabetes, n (%)	1 (4.3)
Dyslipemia, n (%)	1 (4.3)
Smoking habits, n (%)	5 (21.8)
Previous myocardial infarction, n (%)	0 (0)

n: Number of patients

10 patients, with reimplantation of the coronary commissure in 4 and without intervention on the commissure in 6.

In the 5 patients with anomalous origin of the right coronary artery, a right mammary artery graft was performed in 1 case without CPB, 1 patient underwent ostium reimplantation and the unroofing technique was used in 3 (using the modified procedure in the last 2 patients).

Mean CPB time was  $121 \pm 20$  minutes (70-142 min) and mean aortic cross-clamp time was  $73 \pm 11$  minutes (51-94 min).

No postoperative deaths were reported. Patients were hospitalized for an average of  $5.6 \pm 1.1$  days (4 – 8 days) and stayed in the intensive care area for less than 48 hours in all cases. Two patients developed ST-segment deviation associated with pericardial rub suggestive of postoperative pericarditis. One patient presented atrial fibrillation and was treated with magnesium and beta blockers. None of the patients required inotropic support during the postoperative period.

An echocardiogram was performed in the 23 patients 1 day after surgery and before discharge; the studies showed normal coronary blood flow in the neo-ostium, absence of aortic valve regurgitation in patients undergoing unroofing procedure and normal systolic function.

## DISCUSSION

The anomalous origin of the coronary arteries is a rare condition; the most concerning issue related to this abnormality is the risk of myocardial ischemia and sudden death, especially in young asymptomatic athletes. (6, 7)

All our patients were evaluated due to exertion symptoms; the diagnosis of AOCA was not initially suspected and this might have had an influence on the results of few studies.

In this sense, the presence of symptoms is not a rule in the published literature. Eckart et al. reported the presence of prodromal symptoms in only 12 of 39 patients (30%) who died suddenly due to coronary arteries abnormalities, while Angelini reported symptoms in 7% to 45% of patients. Symptoms generally develop during exertion, especially physical activity. (6, 8, 9, 10)

Rest electrocardiogram and even exercise stress test may result normal or nonspecific. All our patients had normal electrocardiograms and the exercise tests were negative in more than 50% of symptomatic patients. (9)

Several studies have reported normal electrocardiograms in patients in whom AOCA was diagnosed and even in those who developed sudden death. Basso et al. studied 12 patients; 9 had normal rest electrocardiograms and all exercise stress tests ( $n = 8$ ) had negative results. (9, 11)

Echocardiography identified the anomalous origin

in 69.6% of cases and the proximal course in 52.2%. The diagnostic value of this test is controversial, with variable results. Several series published in the present decade by Jureidini et al. and Frommelt et al. have given a significant value to the echocardiographic diagnosis of this condition, while Angelini and Flamm criticized and expressed the limitations of the method. These authors pointed out that the incidence of AOCA in the series of Davis et al. using echocardiography was lower (0.1%), compared to studies using coronary angiography (1.1%). Also, in symptomatic patients with syncope who presented sudden death, echocardiography failed to identify the AOCA which was then confirmed by autopsy. Finally, the 1:1 ratio of right to left AOCA reported by echocardiographic series is inconsistent with the 4:1 ratio observed in angiographic series or at autopsies. (12-17)

Transthoracic echocardiography is a simple, inexpensive and noninvasive method which does not use radiations; however, its main limitation is that it is not routinely used for the evaluation of the coronary arteries. For this reason, the operator should have high suspicion about this possible diagnosis. (16)

In all our patients, the diagnosis was confirmed by MSCT angiography, a technique that is considered superior to coronary angiography, the traditional gold standard method, as it is an invasive test that may present diagnostic difficulties and may detect 50% of cases. (18, 19)

MSCT angiography was useful to establish or confirm the anomalous origin and to detect the presence of a proximal interarterial course between the aorta and the pulmonary artery responsible for myocardial ischemia or sudden death. We use the classification of anomalous origin of the coronary arteries that categorizes them in hemodynamically significant or major, potentially associated with risk of sudden death, versus nonsignificant or minor. The former include coronary artery emerging from the pulmonary artery or from the contralateral coronary sinus with an interarterial (between the aorta and the pulmonary artery) or intraarterial (or intramural aortic) course. Retroaortic, prepulmonic and subpulmonary courses are considered minor anomalies. (19-21)

Dynamic compression of an intraarterial or intramural course of the artery is one of the several hypotheses that explain the development of ischemia and sudden death in this population. Table 2 describes the probable mechanisms producing ischemia. (20)

The potential consequences of radiation exposure by this technique have not been clearly determined yet, thus a risk-benefit ratio should be considered. In addition, the method requires the use of contrast agents with the risk of kidney toxicity. (22, 23)

Magnetic resonance angiography has high sensitivity and specificity for the diagnosis of AOCA and can also detect the proximal course of the aberrant vessel; however, we did not use this technique. Among

the limitations to the method, we should mention the impossibility to use it in patients with pacemakers and mechanical heart valve prostheses.

#### Indications for surgery

Despite controversial information about the necessity to treat and how to do it, the surgical approach is mostly used and recommended in symptomatic young patients with AOCA and a proximal interarterial or intramural course.

On the basis of such concepts and findings, all our patients underwent surgery. (10, 24)

When a coronary artery originated from the pulmonary artery, surgery was indicated to avoid myocardial ischemia in the territory of the left circumflex artery due to coronary runoff into the low-pressure pulmonary artery. Coronary reimplantation was used in both cases. This disorder is extremely rare in adults. (25-27)

Three patients underwent mammary artery or saphenous vein grafts; the abnormal ostium was ligated to prevent competitive flow. CPB was used only in one patient. Coronary artery bypass graft surgery is widely known and universally accepted as the treatment of atherosclerotic coronary artery disease; yet, some questions and doubts arise about graft long-term patency in young patients with this anomaly. (28)

Five patients underwent reimplantation of the coronary ostia in the proper sinus under CBP. This technique was complex and demanding in few cases due to a slit like opening of the ostium and the use of autologous pericardial patch. (29)

Unroofing of the coronary artery was performed in 13 patients; the traditional technique was used in 9 patients creating an enlarged neo-orifice with reimplantation of the corresponding aortic commissure, while the modified technique was used in 4 patients, with a limited resection. Table 3 shows the different surgical options with their advantages and limitations. (30-32)

The postoperative outcomes were favorable, as expected in young patients with normal ventricular function and without associated conditions, thus

emphasizing the value of the surgical approach adopted.

#### Study limitations

This is a retrospective evaluation of a very particular series of symptomatic young patients with hemodynamically significant AOCA. We did not compare surgery with other alternative therapies as medical treatment or percutaneous coronary angioplasty with stent implantation. (21, 33, 34)

Therapeutic interventions in asymptomatic patients, older and/or with minor anomalies are still under discussion.

The small number of patients included is another limitation very frequently in the published bibliography, and those studies including a great number of cases are postmortem evaluations. Most of the reports of surgical treatment include a few cases. We strongly agree with the need of creating multicenter registries about this condition.

#### CONCLUSIONS

The defect is more common in the origin of left coronary artery.

1. The diagnostic value of rest electrocardiogram and exercise stress test was low.
2. MSCT angiography was useful to identify the proximal course of the vessel.
3. The unroofing procedure was the surgical technique most used.

**Table 2.** Probable mechanisms producing ischemia

Variables
1. Related with the shape of the coronary ostium of the aberrant vessel (slit like, coronary ostial flap)
2. Related with the proximal course of the vessel (intramural or intraarterial) subjected to extrinsic compression
3. Related with acute angulation of the proximal course of the aberrant vessel
4. Vasospasm
5. Related with possible endothelial lesion secondary to the development of turbulent flow with or without atherosclerosis

**Table 3.** Different surgical techniques; advantages and limitations

Variable	Variable	Limitations
Coronary artery bypass graft	40 years of experience Universally used in this type of patients	Doubts arise about graft long-term patency
Ostium reimplantation	"Physiological" procedure Restores normal anatomy	Technically difficult in occasions Risk of aortic root bleeding
Unroofing	Restores normal anatomy	Risk of aortic valve regurgitation
Modified unroofing	Restores normal anatomy	Prevents risk of aortic valve regurgitation

## RESUMEN

### Diagnóstico y tratamiento quirúrgico del origen anómalo de las arterias coronarias

#### Introducción

El origen anómalo de las arterias coronarias representa una entidad poco frecuente, cuya mayor importancia radica en que se trata de una causa potencialmente prevenible de muerte súbita. Si bien existen controversias respecto de la indicación de tratamiento en general y de cirugía (u otro método de revascularización) en particular, el desarrollo observado en técnicas diagnósticas no invasivas permite un diagnóstico más frecuente y apropiado, lo que nos enfrenta en forma creciente con este tipo de pacientes.

#### Objetivo

Analizar una población de pacientes portadores de origen anómalo coronario a través de la evaluación de la metodología diagnóstica y el tratamiento quirúrgico.

#### Material y métodos

Se evaluaron retrospectivamente pacientes intervenidos entre 2004 y 2010. Se consideraron las características clínicas, la sintomatología, los métodos complementarios, la indicación quirúrgica y las técnicas empleadas.

#### Resultados

Se estudiaron 23 pacientes (17 hombres y 6 mujeres) de entre 18 y 32 años por sintomatología ante esfuerzos, angina en 12 pacientes (52,2%), dolor torácico en 4 casos (17,4%), síncope en 4 (17,4%) y disnea en 3 pacientes (13%). Los electrocardiogramas fueron normales en todos, mientras que la prueba de esfuerzo resultó positiva en 10 casos (43,5%). En todos los pacientes se efectuó un ecocardiograma, que demostró el origen anómalo en 16 (69,5%) e identificó el trayecto proximal en 12 (52,2%). La angiotomografía coronaria efectuó o confirmó el diagnóstico en los 23 pacientes, lo que permitió caracterizar un trayecto intraarterial en los 21 casos con origen desde el seno contralateral. La técnica quirúrgica consistió en el reimplante coronario en 7 casos, en puente (bypass) en 3 y en resección parietal o unroofing en 13 casos. No hubo mortalidad posoperatoria.

#### Conclusiones

La anomalía más frecuente involucró el origen de la coronaria izquierda. El valor diagnóstico de la electrocardiografía fue bajo. La angiotomografía coronaria resultó diagnóstica en todos los casos, lo que permitió caracterizar el trayecto proximal.

El unroofing fue la técnica quirúrgica más empleada.

**Palabras clave >** Anomalías cardiovasculares - Tomografía Cirugía cardíaca

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