# Incidence and treatment of congenital coronary artery anomalies

## Incidence et traitement des anomalies congénitales des artères coronaires

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**RÉSUMÉ**

**Background:** Congenital coronary arteries anomalies are a rare entity. Although their identification started in the 60th, there is a lack of data concerning their frequency and clinical significance in Tunisia.
**Aim:** To characterize clinical and imaging features and mid-term follow up data of congenital coronary artery anomalies in a population of Tunisian adults.
**Methods:** We reviewed the records of 6358 adult patients who underwent coronary angiography between 2009-2015 years in Mongi Slim hospital La Marsa, Tunisia. Multidetector computed tomography was performed on all patients diagnosed having these anomalies and Angelini classification was used for their arrangement. Patients, having intramural coronary artery, were excluded from this study.
**Results:** Thirteen patients had congenital coronary arteries anomalies (seven females and six males). Ten had anomalies of origination and course while the others had anomalies of coronary termination. The right coronary artery was the vessel involved most frequently. It originated from an anomalous coronary ostium in four patients and a unique right coronary artery was reported in one case. An anomalous left main coronary artery was seen in four cases. One patient had the left anterior descending artery originating from the right Valsalva sinus.
Four patients underwent coronary revascularization, one died before the intervention and the remainder received medical management. The mean follow up was 54.1±20 months.
**Conclusion:** Congenital coronary arteries anomalies have a low incidence in adults. Coronary revascularization is actually indicated in anomalous aortic origin with inter aorto-pulmonary course.

**Mots-clés**
Coronary arteries, Congenital anomalies, Angiography.

**SUMMARY**

**Introduction :** Les anomalies congénitales des artères coronaires représentent une entité rare. Bien que leur identification remonte aux années 60, peu d'études tunisiennes ont été publiées sur ce sujet.
**Objectifs :** Rapporter les caractéristiques cliniques et angiographiques de ces anomalies ainsi que les résultats du suivi à moyen terme chez une population d'adultes tunisiens.
**Méthodes :** Nous avons réalisé une étude rétrospective incluant 6358 patients ayant eu une coronarographie à l'hôpital Mongi Slim La Marsa entre 2009 et 2015. Tous les patients présentant ces anomalies ont eu un scanner coronaire. Ceux, avec un pont intra-myocardique, ont été exclus. La classification de ces anomalies a été établie selon celle d'Angelini.
**Résultats :** Treize patients (sept femmes et six hommes) présentaient ces anomalies. Dix avaient des anomalies de naissance ou de trajet alors que trois avaient des anomalies de terminaison. La coronaire droite était le vaisseau le plus impliqué avec quatre cas de naissance à partir d'un ostium anormal et un cas de coronaire droite unique. La coronaire gauche naissait d'un ostium anormal dans quatre cas et un patient avait une artère inter-ventriculaire antérieure naissant du sinus de Valsalva droit. Quatre sujets ont eu une revascularisation coronaire, une patiente est décédéeavant son intervention et les autres ont reçu un traitement médical.La durée moyenne de suivi était de 54,1±20 mois.
Conclusion : L'incidence des anomalies congénitales des artères coronaires est faible. La revascularisation coronaire est indiquée actuellement en cas d'anomalies de naissance avec un trajet inter aorto-pulmonaire.

**Key-words**
Artères coronaires, Anomalies congénitales, Angiographie

## INTRODUCTION

Congenital coronary artery anomalies are a rare entity. They are detected in about 0.16-1.3% of patients undergoing coronary angiography (CA) (1-17). Several classifications were offered. The most used one is proposed by Angelini (18). Although most of them are without clinical significance, some may be responsible for dreadful complications such as sudden death, syncope and acute coronary syndrome. There is a lack of data concerning their frequency and clinical significance in Tunisia. We report our experience through a population of 6358 patients.

## METHODS

We reviewed the records of 6358 adult patients who underwent CA between 2009-2015 years in Mongi Slim Hospital La Marsa, Tunisia. Angiograms were reviewed by at least two experienced independent observers, who reached a consensus on the origin and course of the anomalous coronary artery. Sixty-four-slice Multidetector coronary computed tomography angiogram (CCTA) was performed on all patients diagnosed having these anomalies and Angelini classification was used for their arrangement (18). Patients, having intramural coronary artery and those with coronary anomalies occurring as part of a complex congenital heart disease, were excluded from this study.

## RESULTS

**Study population:**

Among 6358 adult patients (mean age, 65.2±19 years [range, 21—85 years]) who underwent diagnostic CA, 13 (0.20%) patients (seven females and six males) had congenital coronary artery anomalies. Their mean age was 51.3±11 years. The indication for angiography was the evaluation of coronary artery disease (two patients with acute coronary syndrome and nine with stable angina) except for two patients (dilated cardiomyopathy). The clinical and angiographic characteristics of the patients are presented in Table 1.

**Table 1:** Clinical and imaging characteristics of patients with congenital coronary arteries anomalies

| Case | Sex/Age (years) | Presentation | Coronary arteries anomalies | Atherosclerotic lesions | Treatment |
|---|---|---|---|---|---|
| 1 | H/54 | ACS | RCA originating from a high ostium | TS LAD-2nd Dg | PTCA LAD-2nd Dg |
| 2 | F/52 | ACS | LMCA originating from the RSV | TS LMCT-LAD-RCA | Patient deceased |
| 3 | F/48 | Stable angina | RCA originating from the LSV | - | Medical |
| 4 | F/55 | Stable angina | LMCT absent | - | Medical |
| 5 | H/48 | Stable angina | LAD originating from the RSV | Occlusion of the RCA | CABG CD-IVA |
| 6 | F/47 | Stable angina | LMCA originating from the RSV | - | Medical |
| 7 | H/52 | Stable angina | RCA originating from the LSV | TS LMCT-LAD-CXA | CABG LAD-CXA-RCA |
| 8 | F/42 | Stable angina | LMCA originating from the RSV | - | Medical |
| 9 | H/60 | Stable angina | RCA originating from the LSV | TS LAD | CABG LAD-RCA |
| 10 | F/55 | DCM | Unique RCA | - | Medical |
| 11 | H/32 | DCM | Coronary-to-pulmonary fistula | - | Medical |
| 12 | H/60 | Stable angina | Coronary-to-LV and LA fistula | - | Medical |
| 13 | F/62 | Stable angina | Coronary-to-LV and LA fistula | - | Medical |

ACS: acute coronary syndrome, CABG: coronary artery bypass graft, CXA: circumflex artery, DCM: dilated cardiomyopathy, Dg: diagonal artery, LAD: left anterior descending artery, LMCA: left main coronary artery, LMCT: left main coronary trunk, LA: left atrium, LSV: left sinus of Valsalva, LV: left ventricle, PTCA: percutaneous transluminal coronary angioplasty, RCA: right coronary artery, RSV: right sinus of Valsalva, TS: tight stenosis.

**Congenital coronary artery anomalies:**

Ten patients had anomalies of origination and course while the other ones had anomalies of coronary termination.

The right coronary artery (RCA) was the vessel involved most frequently (seven cases). In fact, it originated from the left sinus of Valsalva in three patients [separately from the left main coronary artery (LMCA) in two cases and from the same ostium in one case] and among them, in two patients, it had inter-aortopulmonary course (figure1). In another patient, it originated from an ostium highly located from the right sinus of Valsalva (RSV) and lateralized to the left. A unique RCA was reported in one case (figure2). The LMCA originated separately from the RSV in three patients and there was no left coronary main trunk (split origination of the LMCA) in one case. The left anterior descending artery originated from the RSV with inter-aortopulmonary course in one patient (figure 6).

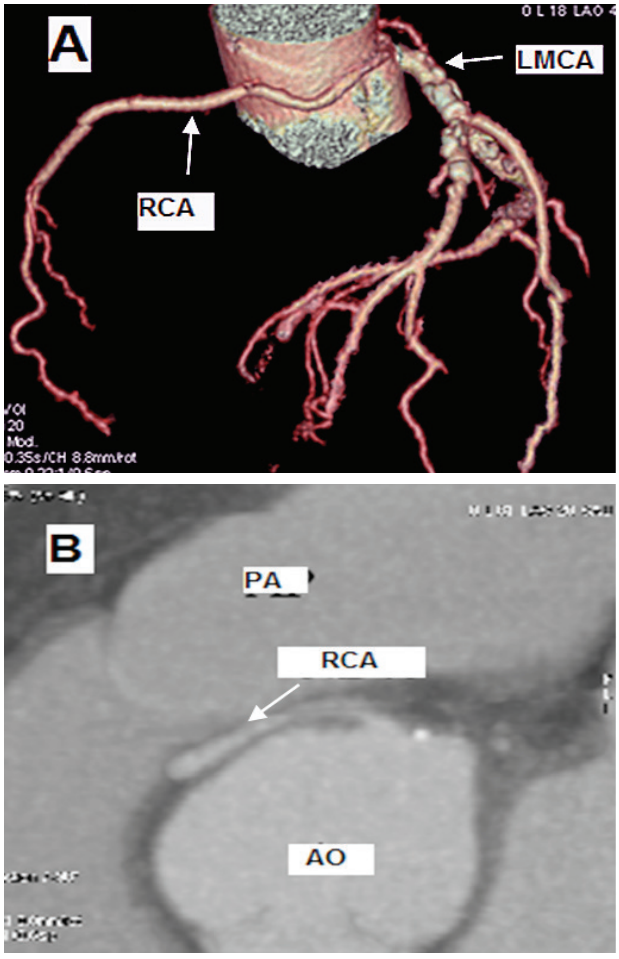


**Figure 1:** CCTA, 3D reconstruction volume rendering (A) and 3D curved planar reconstruction (B): anomalous RCA originating from the LSV with inter-aortopulmonary course (case 7).

CCTA: coronary computed tomography angiogram, RCA: right coronary artery, LSV: left sinus of Valsalva, PA: pulmonary artery, AO: aorta.

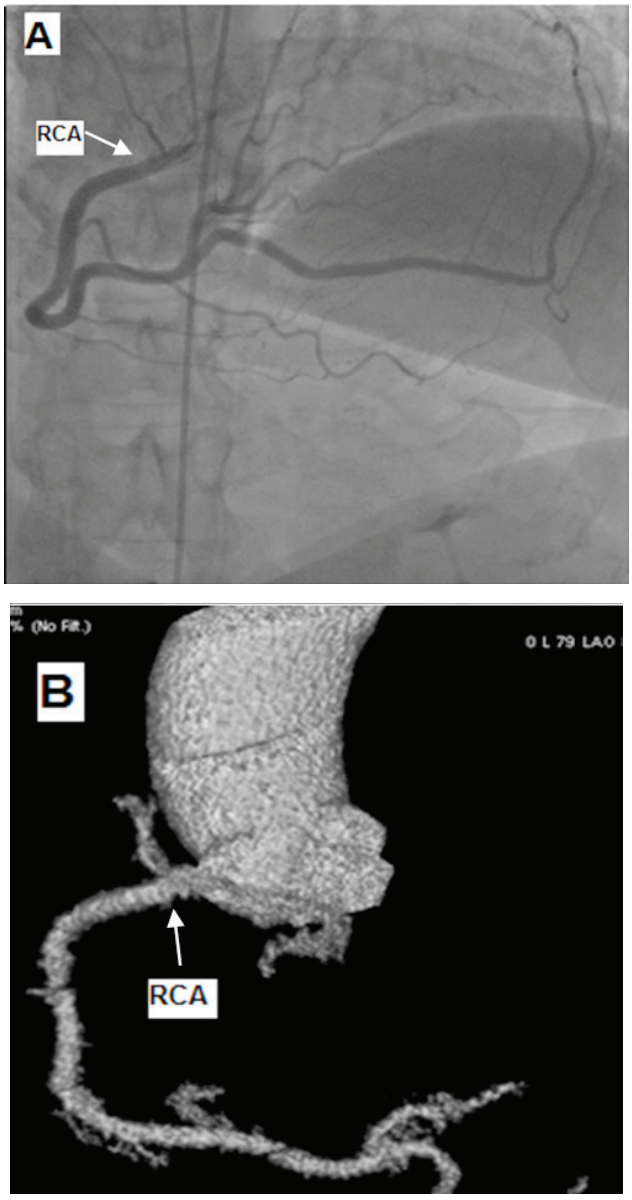


**Figure 2: (**A) coronary angiography, left profile view: dominant RCA and the PDA seeming to be extended by the LAD. (B) CCTA, 3D reconstruction volume rendering: LMCA absent and unique RCA (case 10).

RCA: right coronary artery, PDA: posterior descending artery, LAD: left anterior descending artery, CCTA: coronary computed tomography angiogram, LMCA: left main coronary artery, RCA: right coronary artery.

The left anterior descending artery originated from the RSV with inter-aortopulmonary course in one patient (figure 3).
Three cases of coronary fistulas were reported: a case of a fistula from the LMCA to the main pulmonary artery without a significant shunt and two cases of fistulas from the RCA to the left atrium and ventricle.
In all these 13 cases, using multidetector CCTA was primary to assess with certainty the course of the anomalous artery.

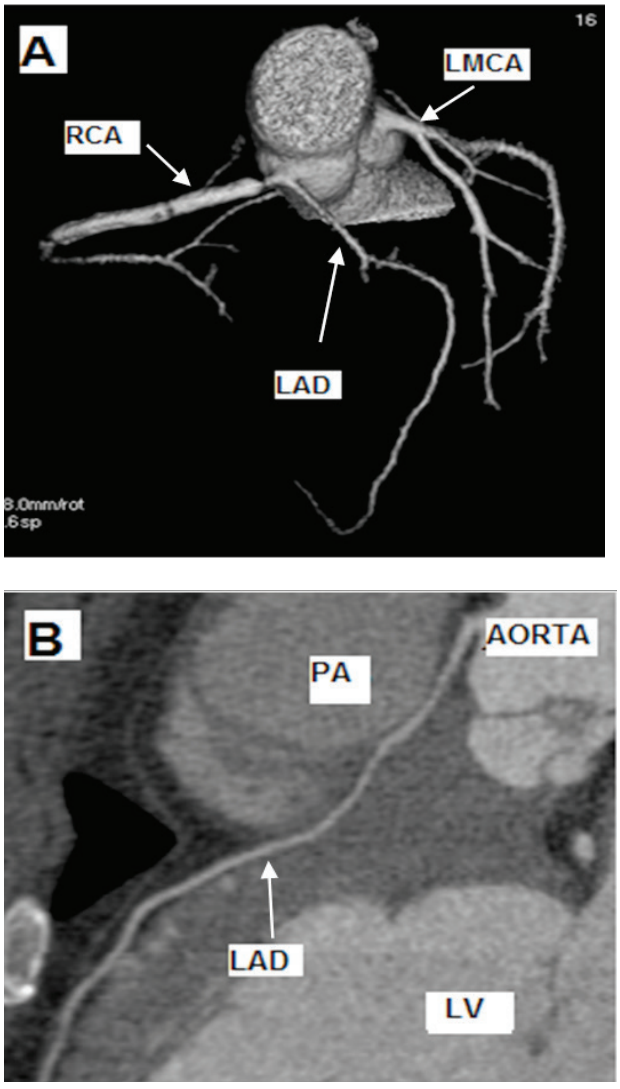


**Figure 3:** CCTA reconstructions, 3D rendering volume (A) and 2D curved planar (B). The LAD arises from the RCA with interarterial course before crossing through the inter-ventricular groove (case 5).
CCTA: coronary computed tomography angiogram, LAD: left anterior descending artery, RCA: right coronary artery, PA: pulmonary artery, LV: left ventricle.

Atherosclerotic lesions were found in five patients and only two among them had significant lesions on the anomalous coronary artery. Four patients underwent coronary revascularization. One had percutaneous transluminal coronary angioplasty (PTCA) and the other three had coronary artery bypass graft (CABG). One patient died before the intervention (CABG) due to ventricular arrhythmia and the remainder received medical management.
These patients are asymptomatic since intervention with a mean follow up of 54.1±20 months.

## DISCUSSION

**Incidence:**
Congenital coronary arteries anomalies are a rare entity. Some retrospective angiographic studies have been published, assessing the incidence of different congenital coronary arteries between 0.16% and 0.31% of patients undergoing coronary angiography as shown in Table 2.

**Table 2:** Incidence of congenital coronary anomalies in patients undergoing coronary angiography

| Authors | Year of publication | Number of patients | Number of anomalies | Angiographic incidence (%) |
|---|---|---|---|---|
| **Engel et al (7)** | 1975 | 4250 | 51 | 1.2 |
| **Wilkins et al (15)** | 1988 | 10661 | 83 | 0.78 |
| **Yamanaka et al (16)** | 1990 | 126 595 | 1672 | 1.3 |
| **Topaz et al (13)** | 1992 | 13010 | 57 | 0.44 |
| **Kaku et al (10)** | 1996 | 17731 | 56 | 0.31 |
| **Barriales Villa et al (3)** | 2001 | 13500 | 54 | 0.4 |
| **Aydinyar et al (1)** | 2005 | 12059 | 113 | 0.7 |
| **Ouali et al (17)** | 2009 | 7330 | 20 | 0.27 |
| **Present study** | 2016 | 6358 | 13 | 0.20 |

The overall incidence of congenital coronary artery anomalies in our population was in agreement with the incidence reported in other series (0.16-1.3%) (1-17).

However, it may even be higher if detected by multi-slice coronary computed angiography. We did not include patients with muscular bridge (intramural coronary artery) or with a congenital heart disease which could increase the overall incidence. To the best of our knowledge, this is the second report in Tunisia (17). More women than men had anomalous coronary arteries in contradiction with other series (8,17).

While many series showed a higher incidence of anomalous coronary arteries affecting the RCA, others showed their predilection for the LMCA or even the circumflex artery (1,3,4,8,10,12,13,16,17,19). In our population, the RCA was the vessel involved most frequently. Nonetheless, anomalies of origination and course, which were the most frequent ones (1-3,18), involved specially and equally the RCA and the LCMA. No case of anomalous circumflex artery was reported in our report. This discrepancy may be explained by genetic and geographic features between different populations but also inside the same population.

**Congenital coronary artery anomalies and atherosclerotic lesions:**

Involvement of anomalous coronary arteries in atherosclerotic disease is at best controversial. Many reports didn't show an increased risk for development of atherosclerotic lesions within anomalous coronary arteries (8,13,17). Ouali et al found that only 20% of their patients had evidence of severe atherosclerotic lesions within the anomalous coronary arteries and that none of these lesions was responsible for ischemia manifestation (17). Our study is in agreement with others in that there is no predisposition for atherosclerotic involvement of the anomalous vessels (8,13,17).

**Advantages of coronary computed tomology angiogram:**

Although CA is, to date, the gold standard for the diagnosis of coronary disease, it fails repeatedly to highlight these anomalous coronary arteries and especially the analysis of the ectopic artery course. In addition to its noninvasive trait, recent advances in technology renders CCTA the method of choice for screening for detecting coronary artery anomalies as recommended by the American college of cardiology (class I, level B) and makes up for the shortcomings of CA in many ways (20).

In fact, the prevalence of anomalous coronary arteries seems to be higher when detected by CCTA (23,24). Studies reported that CA could identify only between 31% and 55% of these anomalies when compared to CCTA (24-27). Moreover, it specifies the course of the ectopic coronary artery and its relationship to adjacent structures including vascular ones.

Reconstructions allow better study of the relationship between the aberrant ostium and that of the normal coronary artery (21-24).

**Treatment strategy:**

The majority of congenital coronary artery anomalies (81%) have no clinical impact. It is now admitted that "malicious" forms with risk of sudden death are those with interarterial courses (Table 3) (28-30). Eckart et al reported that 33% of sudden cardiac deaths in young patients are secondary to implantation of the coronary artery on the opposite sinus with interarterial course (28-31).

**Table 3**: Anomalous coronary artery course in relation with the great vessels

| | |
|---|---|
| **Type A** | Coronary artery passing in front of the pulmonary artery (prepulmonary course) |
| **Type B** | Coronary artery passing between the aorta and the pulmonary artery (interarterial course) |
| **Type C** | Coronary artery passing through the infundibular septum (intraseptal course) |
| **Type D** | Coronary artery passing behind the aorta (retroaortic course) |

Angellini reported that these interarterial courses are more frequent in case of abnormal origin of the RCA, which is in agreement with our observation (18).

The most frequently mentioned mechanism is a pressure increase and expansion in the aorta and the pulmonary artery during exercise, causing a compression of the coronary having an aberrant course between these two vessels with myocardial ischemia (32). The second hypothesis advanced to explain the myocardial ischemia is the convoluted course (kinking) of the coronary artery passing between the aorta and pulmonary artery (33). We report in this study three cases of anomalous aortic origins of the coronary arteries with interarterial course.

The prognosis of these "malicious" anomalous coronary arteries is good if their treatment is performed early. For a long time, given the difficulty of estimating the risk of

sudden death and poor results in the medium and long-term outcome of CABG, no therapeutic consensus is validated. However, after several recent studies, the American College of Cardiology advocate coronary revascularization if the LMCA originated from the RSV with interarterial course or if the RCA originated from the left sinus of Valsalva with interarterial course and with evidence of myocardial ischemia (class I, level B) (18,20,28-30,34-37).

Revascularization may be considered in case of abnormal rise of the LAD with interarterial course (class IIb, level C). The revascularization is mainly surgical and recently by PTCA (20,37).

Surgical methods include CABG, ostial reimplantation, the unroofing technique and pulmonary artery translocation (38,39). CABG is preferred when there is associated atherosclerotic obstructive coronary artery disease. Although it is, to date, the most recommended method; it has inherent limitations with limited graft patency (40). Unroofing and translocation procedures are appropriate and effective for certain morphological types, with both techniques showing favorable outcome at mid-term follow-up (38,39).

Unfortunately, aortic insufficiency can result even late after unroofing and myocardial ischemia may persist particularly in patients with repaired coronary artery with interarterial course (41,42).

PTCA seems technically challenging due to difficulties in selective cannulation of the aberrant ostium, the peculiar anatomical milieu, type of stent and ideal degree of stent dilation (43-45).

The transradial approach is as good as transfemoral approach (43). PTCA is feasible with reasonable amount of contrast and radiation and high success rate (45-48). However, these results are limited to anecdotal cases reports and few series (45,47-48).

In our study, three patients with these “malicious” anomalies had in addition severe atherosclerotic lesions and the indication of a CABG seemed logical. These patients are asymptomatic since surgery with a mean follow up of four years.

## CONCLUSION

Congenital coronary artery anomalies have a low incidence in adults. Although they represent a rare entity and that the majority of these anomalies are without clinical significance, their identification is crucial due to the existence of “malicious” anomalies with fatal impact.

Coronary revascularization is currently indicated in cases of anomalous origin with interarterial course. It is based essentially on CABG with a growing interest in PTCA.

**Disclosures:**

No conflict of interests.

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