Anomalous Origin of the Coronary Artery from the Pulmonary Artery in Children and Adults: A Pictorial Review of Cardiac Imaging Findings

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Anomalous origin of the coronary artery from the pulmonary artery is a rare and potentially fatal congenital heart defect. Up to 90% of infants with an anomaly involving the left coronary artery die within the first year of life if left untreated. Patients who survive beyond infancy are at risk of sudden cardiac death. Cardiac CT and MRI are increasingly being used for the accurate diagnosis of this anomaly for prompt surgical restoration of the dual coronary artery system. Moreover, life-long imaging surveillance after surgery is necessary for these patients. In this pictorial review, multimodal cardiac imaging findings of this rare and potentially fatal coronary artery anomaly are comprehensively discussed, and representative images are provided to facilitate the understanding of this anomaly.

Keywords: Cardiac catheterization; Cardiac computed tomography; Cardiac magnetic resonance imaging; Coronary artery anomaly; Echocardiography

INTRODUCTION

Anomalous origin of the left or right coronary artery from the pulmonary artery is a rare and hemodynamically significant coronary anomaly seen in 1 of 300000 live births and accounts for 0.25–0.5% of all congenital heart defects [1-3]. The anomaly was first discovered during an autopsy in 1865 [4] and was later described as a clinical syndrome (known as Bland-White-Garland syndrome) in a 3-month-old infant in 1933 [5]. The anomalies are typically divided into infant and adult types [3]. Infants with an anomaly of the left coronary artery present with myocardial infarction and congestive heart failure because of the gradual decrease in pulmonary arterial pressure at approximately two months of life and poorly developed inter-coronary collateral arteries. Up to 90% of these infants die within the first year of life if left untreated [6]. This anomaly involving the left coronary artery rarely manifests in adults but is an important cause of sudden cardiac death [3,7]. The development of adequate inter-coronary collateral flow supplying the left ventricle delays the manifestations of the anomaly in adulthood. Coronary artery dominance is a determinant of the extent of myocardial ischemia in this anomaly [8]. Notably, the increased use of modern non-invasive cardiac imaging modalities, such as CT and MRI, has substantially increased the number of adults detected with this anomaly [7]. Compared to the anomalies involving the left coronary artery, those involving the right coronary artery are rarer and less severe [9,10]. The higher incidence of the anomalies involving the left coronary artery may be explained by the proximity of the left coronary bud to the pulmonary artery sinus [9,10]. In addition, myocardial ischemia in the anomaly involving the right coronary artery is less prominent because the oxygen demand of the right ventricle is less than that of the left ventricle [10]. Other cardiac anomalies, such as atrial septal defect, ventricular septal defect, and coarctation of the aorta,
might coexist in approximately 5% of the cases with an anomalous origin of the left coronary artery from the pulmonary artery [3]. The aortopulmonary window is commonly associated with approximately 24% of the cases with an anomalous origin of the right coronary artery from the pulmonary artery [9]. The diagnosis of this anomaly has been made using various imaging modalities, such as cardiac catheterization, echocardiography, cardiac CT, and cardiac MRI. Prompt and accurate diagnosis is critical for timely surgical management, such as aortic reimplantation of the anomalous coronary artery, Takeuchi procedure, and placement of a coronary artery bypass graft combined with ligation of the anomalous coronary artery origin, to restore a two-coronary-artery circulatory system have shown excellent results [3,7,9]. It is difficult to obtain comprehensive and updated medical knowledge about this anomaly because of its rarity; however, cardiac imagers might be able to make a correct diagnosis if they are familiar with its characteristic imaging findings. This pictorial review describes the cardiac imaging findings of anomalous origin of the left or right coronary artery from the pulmonary artery in children and adults (Table 1).

### Hemodynamic Alterations of Anomalous Origin of the Coronary Artery from the Pulmonary Artery

In fetal and early neonatal periods, antegrade flow in the affected coronary artery anomalously arising from the pulmonary artery is maintained because pulmonary arterial pressure equals systemic pressure. However, as pulmonary vascular resistance subsequently drops and the ductus arteriosus is closed, the pulmonary arterial pressure gradually decreases, which leads to the development of decreased antegrade and eventually reversed flow in the affected coronary artery draining into the pulmonary artery. As a result, the so-called “coronary steal” phenomenon driven by the pressure difference between systemic and pulmonary arterial systems occurs. Hemodynamic alteration results in not only a left-to-right shunt but also, more importantly, myocardial ischemia and infarction, unless inter-coronary collateral circulation is sufficiently developed. In fact, the extent of the acquired inter-coronary collaterals determines the extent of the myocardial ischemia. Therefore, adult patients almost always demonstrate intercoronary collaterals on cardiac imaging.

### Table 1. Cardiac Imaging Findings of Anomalous Origin of the Coronary Artery from the Pulmonary Artery in Children and Adults

| Imaging Modality          | Infant Type                                                                 | Adult Type                                                                 |
|---------------------------|-----------------------------------------------------------------------------|---------------------------------------------------------------------------|
| Chest radiography         | Cardiomegaly with or without mild pulmonary edema                           | No or mild cardiomegaly                                                  |
| Echocardiography          | Anomalous connection between the affected coronary artery and the pulmonary artery; reversed flow in the affected coronary artery; no inter-coronary collaterals; mitral regurgitation; myocardial infarction; ventricular enlargement with impaired function | Anomalous connection between the affected coronary artery and the pulmonary artery; reversed flow in the affected coronary artery; dilated inter-coronary collaterals; no or mild mitral regurgitation; no or mild ventricular enlargement; no or mild ventricular hypertrophy |
| Cardiac CT                | Anomalous connection (including intramural route) between the affected coronary artery and the pulmonary artery; no inter-coronary collaterals; myocardial infarction; ventricular enlargement with impaired function | Anomalous connection between the affected coronary artery and the pulmonary artery; dilated inter-coronary and extra-cardia collaterals; no or mild ventricular enlargement; no or mild ventricular hypertrophy |
| Cardiac MRI               | Not recommended due to limited spatial resolution and long examination time | Anomalous connection between the affected coronary artery and the pulmonary artery; reversed flow in the affected coronary artery; dilated inter-coronary collaterals; no or mild mitral regurgitation; myocardial perfusion deficit; myocardial infarction; myocardial fibrosis; no or mild ventricular enlargement; no or mild ventricular hypertrophy |
| Stress-rest myocardial perfusion single-photon emission CT | Fixed or reversible myocardial perfusion defect | Fixed or reversible myocardial perfusion defect |
Nevertheless, collateral flow is likely insufficient to supply the subendocardial region. The region with chronic ischemia typically acts as a substrate for malignant ventricular dysrhythmia, which increases the risk of sudden cardiac death in adult patients.

**Chest Radiography**

In the infant type, initial chest radiography showed cardiomegaly due to the enlargement of the left cardiac chambers with or without mild pulmonary edema (Fig. 1) [11]. In contrast, initial chest radiography showed no or mild cardiomegaly in adults. Although it might reveal the presence and severity of cardiomegaly and congestive heart failure, chest radiography is nonspecific. Therefore, other cardiac imaging modalities are necessary to definitively diagnose the anomalous origin of the coronary artery from the pulmonary artery.

**Cardiac Catheterization**

Cardiac catheter angiography was used for the diagnosis of anomalous origin of the coronary artery from the pulmonary artery (Fig. 2). The average year of the publications where the diagnosis was made using cardiac catheter angiography was 2001 in a systematic review [9], which means that the imaging modality is out-of-date. Preoperative cardiac catheterization can provide an accurate diagnosis and allow assessment of the degree of inter-coronary collateral flow, left-to-right shunt fraction, and left ventricular end-diastolic and pulmonary arterial pressures [9,12]. However, due to a low but well-known risk of procedure-related complications, especially in young children [13], and the availability of non-invasive cross-sectional cardiac imaging modalities, diagnostic catheter angiography is no longer the preferred modality.

**Echocardiography**

Transthoracic echocardiography is the primary diagnostic imaging modality for this anomaly, and it can demonstrate an anomalous connection between the affected coronary artery and the pulmonary artery, abnormal flow in the affected coronary artery and the adjacent pulmonary artery, intercoronary collateral flow, degree of mitral regurgitation, extent of myocardial infarction, and left ventricular function (Fig. 3) [14,15]. However, the diagnostic accuracy of echocardiography for this anomaly is not absolute and has been reported to be in the range of 46.0–80.0%; it is relatively lower in adult patients than in pediatric patients [7,11,15,16]. Missed or false-negative cases on echocardiography might be attributed...
to invisible echogenicity of the lateral aortic and arterial walls superimposed by the transverse sinus, which might be misinterpreted as a normal connection between the coronary artery and the aorta [17]. Therefore, an additional cardiac imaging study, such as CT or MRI, is usually necessary to make an accurate diagnosis. However, a recent

Fig. 2. Preoperative cardiac catheterization and angiography of anomalous origin of the left coronary artery from the pulmonary artery.
A. Frontal selective right coronary artery angiography of the infant type demonstrates faint visualization of the left coronary artery draining into the main pulmonary artery (arrow). Notably, the inter-coronary collateral vessels are poorly developed. B. Frontal selective right coronary artery angiography of the adult type shows the dilated, tortuous right and left coronary arteries with well-developed inter-coronary collateral arteries. The origin of the left coronary artery is anomalously connected to the main pulmonary artery (arrows). C. Frontal pulmonary angiography reveals regurgitant flow into the left coronary artery (arrows) in the infant type of this anomaly.

Fig. 3. Preoperative transthoracic echocardiography of anomalous origin of the left coronary artery from the pulmonary artery.
A. A parasternal echocardiographic image shows an anomalous connection (arrow) of the left coronary artery to the lateral aspect of the proximal MPA in the infant type. B. A Doppler echocardiographic image reveals reversed flow (red color) in the left circumflex artery (arrows) in the infant type. C. Apical four-chamber echocardiographic image demonstrates characteristic inter-coronary collateral arteries (arrows) in the interventricular septum in the adult type. D, E. Apical four-chamber echocardiographic images showing the thinned, echogenic anterolateral papillary muscle (arrows in D) with ischemic damage-induced fibrosis and calcification. In addition, Doppler echocardiography (E) shows mitral regurgitation (red color). Of note, the LA and LV are enlarged. AA = ascending aorta, LA = left atrium, LV = left ventricle, MPA = main pulmonary artery, RV = right ventricle
case report demonstrated that pulmonary artery-focused contrast echocardiography with supplemental oxygen improved the diagnosis of anomalous origin of the left coronary artery from the pulmonary artery in an infant with suspected dilated cardiomyopathy [18].

Cardiac CT

Coronary artery visibility on cardiac CT has dramatically increased with the use of electrocardiography-synchronized scanning, even in normally breathing young children [19-
Fig. 5. Postoperative cardiac CT after reimplantation of the left coronary artery for treating anomalous origin of the left coronary artery from the pulmonary artery.
A. An oblique sagittal volume-rendered CT image shows the patent left coronary artery (arrow) reimplanted into the aorta in the infant type.
B. An oblique frontal volume-rendered CT image reveals an anastomotic stenosis (arrows) of the reimplanted left coronary artery in the adult type. The coronary arteries remain dilated and tortuous. LA = left atrium, LV = left ventricle, RV = right ventricle.

Fig. 6. Cardiac MRI of anomalous origin of the left coronary artery from the pulmonary artery.
A, B. Short-axis cine images show an anomalous connection (arrow in A, B) of the left coronary artery to the MPA with dark intensity due to the turbulent flow (arrow in B). C. Series of short-axis cine images used for ventricular function assessment. For ventricular volumetry, a simplified contouring method is usually used. D, E. Short-axis (D) and long-axis (E) late gadolinium enhancement images demonstrate subendocardial myocardial infarction in the anteroseptal wall of the LV (arrows). AA = ascending aorta, LA = left atrium, LV = left ventricle, MPA = main pulmonary artery, RA = right atrium, RV = right ventricle.
Anomalous Origin of the Coronary Artery from the Pulmonary Artery

Owing to the improved coronary artery visibility, CT is now considered the diagnostic imaging modality of choice for evaluating coronary artery anomalies [22-25]. This is also true for the preoperative diagnosis of anomalous origin of the coronary artery from the pulmonary artery [11,16,26,27]. In addition, myocardial infarction can be diagnosed using late iodine enhancement CT acquired 6-15 minutes after the intravenous administration of an iodinated contrast agent [28,29], and ventricular function can be assessed using data acquired during the end-systolic and end-diastolic phases [30,31]. Preoperative cardiac CT showed an anomalous connection between the affected coronary artery and the pulmonary artery, inter-coronary collateral flow, extent of myocardial infarction, and left ventricular function (Fig. 4).

The anomalous origin commonly arises from the facing pulmonary sinuses; however, it may rarely originate from the distal main pulmonary artery and even from the right branch pulmonary artery. If the anomalous origin is close to the aorta, coronary button transfer is adequate. However, other surgical techniques, such as the Takeuchi procedure and the use of a rolled conduit, should be considered in cases with an inadequate length of the intact anomalous coronary artery [3,16,32]. In a rare variant with an intramural aortic route, cardiac CT shows a slit-like anomalous vessel arising from the superomedial aspect of the distal pulmonary artery and running between the pulmonary trunk and the ascending aorta. Therefore, coronary artery unroofing based on CT findings may be planned and performed [16,27]. In the adult type, extracardiac collateral circulation, such as the dilated bronchial arteries, might be detected in addition to the inter-coronary collateral arteries [7]. Cardiac CT is also useful in evaluating the patency of the repaired coronary artery and postoperative complications such as stenosis at the reimplantation site (Fig. 5), supravalvular pulmonary stenosis, baffle leak after Takeuchi repair, and stenosis and occlusion of the coronary bypass graft [33-35].

**Cardiac MRI**

Cardiac MRI can be used for comprehensive evaluation using coronary angiography, vascular and valvular flow imaging, cine imaging for ventricular function assessment, myocardial perfusion imaging, and late gadolinium enhancement imaging (Fig. 6) [3,12,36]. However, coronary MR angiography is usually inadequate at delineating small coronary arteries in pediatric patients [37,38]. For such cases, CT is the best imaging modality for diagnosis as well.

**Fig. 7. Stress-rest myocardial perfusion single photon emission CT of anomalous origin of the left coronary artery from the pulmonary artery.**

A. Radionuclide myocardial perfusion study shows a large fixed perfusion defect in the anteroseptal wall of the LV. B. Radionuclide myocardial perfusion study shows a large partially reversible perfusion defect in the anteroseptal wall of the LV. LV = left ventricle
as for early postoperative follow-up to identify the early postoperative complications. In contrast, cardiac MRI is recommended as a long-term postoperative surveillance tool due to its ability to comprehensively evaluate myocardial viability [3,12,36]. Recently, four-dimensional flow MRI has been used to visualize the retrograde flow from the left anterior descending artery into the main pulmonary artery in a 10-year-old girl with the anomaly [39]. Cardiac T1 mapping was recently reported to demonstrate globally elevated non-contrast T1 (1117 ms) and extracellular volume fraction (> 32.4%) values in infants and adults, indicating the presence of diffuse myocardial fibrosis [40,41]. However, future studies are warranted to determine the clinical significance of cardiac T1 mapping.

**Myocardial Perfusion SPECT**

Stress-rest myocardial perfusion single-photon emission CT can be used to determine the hemodynamic significance of coronary arteries with an anomalous origin or course by identifying reversible myocardial perfusion defects [42]. It can also be used to identify ischemic but viable myocardium as well as irreversibly damaged myocardium in this anomaly before and after surgical management (Fig. 7) [43,44].

**CONCLUSION**

Prompt and accurate diagnosis of anomalous origin of the coronary artery from the pulmonary artery is critical for planning and performing optimal surgical treatment to restore the dual-coronary-artery system. Cardiac CT and MRI play a pivotal role in providing detailed presurgical information and ascertaining postsurgical functional improvement in both pediatric and adult patients with this anomaly. Cardiac imagers should be familiar with the multimodal imaging findings of this rare and potentially fatal anomaly.

**Conflicts of Interest**
The author has no potential conflicts of interest to disclose.

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