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Imaging Findings of Coronary Artery Fistula in Children: A Pictorial Review

Hyun Woo Goo

Department of Radiology and Research Institute of Radiology, Asan Medical Center, University of Ulsan College of Medicine, Seoul, Korea

Coronary artery fistula, defined as an abnormal communication between the coronary arteries and a cardiac chamber (most commonly) or a thoracic great vessel, may result in hemodynamically significant problems due to vascular shunting in children. Echocardiography, cardiac catheterization, cardiac MRI, and cardiac CT may be used to evaluate coronary artery fistula in children. Recently, CT has played a pivotal role for the accurate diagnosis of coronary artery fistula in children. Surgical or interventional treatment is performed for hemodynamically significant coronary artery fistulas. In this pictorial review, the detailed imaging findings of coronary artery fistula in children are described.

Keywords: *Cardiac catheterization; Children; Cardiac CT; Coronary artery anomalies; Echocardiography*

INTRODUCTION

Coronary artery fistula, defined as a rare congenital or acquired abnormal communication between the coronary arteries and a cardiac chamber or a thoracic great vessel without an intervening capillary network, was first described in 1865 by Krause [1]. The prevalence of coronary artery fistula is estimated to be 0.002% in the general population, and has been identified in 0.05%–0.25% of patients who underwent catheter coronary angiography [2,3]. More recently, the reported prevalence of coronary artery fistula determined with coronary CT angiography has been relatively high, with a range of 0.19%–0.91% [4-6].

Congenital coronary artery fistulas are more common (> 90%) than acquired ones [7]. Based on the drainage site of the fistula, the anomaly can be classified as coronary cameral fistula (draining into a cardiac chamber), coronary-to-pulmonary artery fistula, coronary artery-to-coronary sinus (or any other cardiac vein) fistula, and coronary artery-to-bronchial artery fistula [7]. Among them, coronary cameral fistula is the most common type detected on coronary angiography [7]. In adults, coronary-to-pulmonary artery fistula has been increasingly detected with the increased use of coronary CT angiography, and is currently the most common coronary artery fistula type, accounting for 76.8%–89.5% of cases [4,8,9]. In contrast, coronary-to-pulmonary artery fistula is rare in children, and only 7.5% of patients with coronary-to-pulmonary artery fistula were younger than 18 years old [5].

Coronary artery fistula occurs in isolation in 55.0%–88.0% of cases, whereas 5.0%–30.0% of cases are associated with other congenital heart diseases [10]. Associated congenital defects include atrial septal defect, ventricular septal defect, tetralogy of Fallot, patent ductus arteriosus, and pulmonary atresia with an intact ventricular septum [11].

Imaging findings of coronary artery fistula in adults have been frequently described in the literature [3,4,6-9,12-14], while imaging findings of coronary artery fistula in children have been rarely reported [5,15]. Therefore, this paper aimed to illustrate the detailed imaging findings of coronary artery fistula in children.

Clinical Scenario and Hemodynamic Significance

Unlike adult patients with coronary artery fistula who are

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Corresponding author: Hyun Woo Goo, MD, PhD, Department of Radiology and Research Institute of Radiology, Asan Medical Center, University of Ulsan College of Medicine, 88 Olympic-ro 43-gil, Songpa-gu, Seoul 05505, Korea.
• E-mail: ghw68@hanmail.net

Korean Journal of Radiology
KJR

usually asymptomatic, the majority of pediatric patients are symptomatic, as children commonly develop large fistulous connections [15]. The most common clinical presentation of coronary artery fistula in children is a continuous murmur. With increasing patient age, the fistula gradually enlarges with increased shunt flow via the fistula. In contrast, spontaneous closure of the fistula is rare, accounting for 1.0%–2.0% of cases [7]. Aneurysmal changes are frequently seen in the distal segments of coronary artery fistulas (24.2%–87.1%) [4,6,9,12,13]. The amount of shunt flow depends on the size of the fistula, the presence of stenosis, and the pressure level at the drainage site.

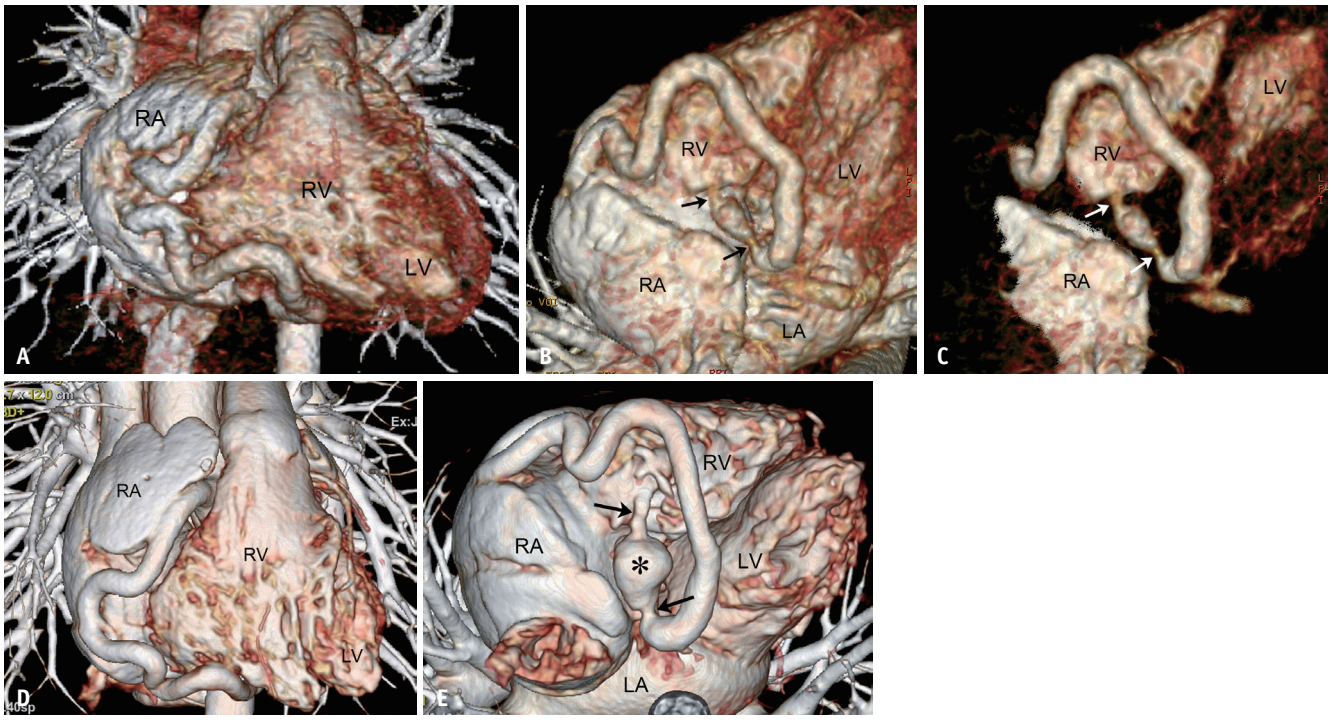

Fig. 1. Coronary cameral fistula between the right coronary artery and the RV.
A. Initial frontal volume-rendered CT image showing the diffusely dilated right coronary artery in a 5-month-old boy. **B, C.** Initial inferior volume-rendered CT images with a full slab **(B)** and with a thinner slab **(C)** demonstrating the single drainage site of the fistula into the inferomedial basal portion of the RV. Two narrowings (arrows) are noted at the drainage site. **D, E.** Follow-up frontal **(D)** and inferior **(E)** volume-rendered CT images performed 8 years later showing increased dilatation of the right coronary artery and the development of aneurysmal change (asterisk) at the drainage site. The two narrowings (arrows) showed interval increase in diameter due to increased left-to-right shunt through the fistula. LA = left atrium, LV = left ventricle, RA = right atrium, RV = right ventricle

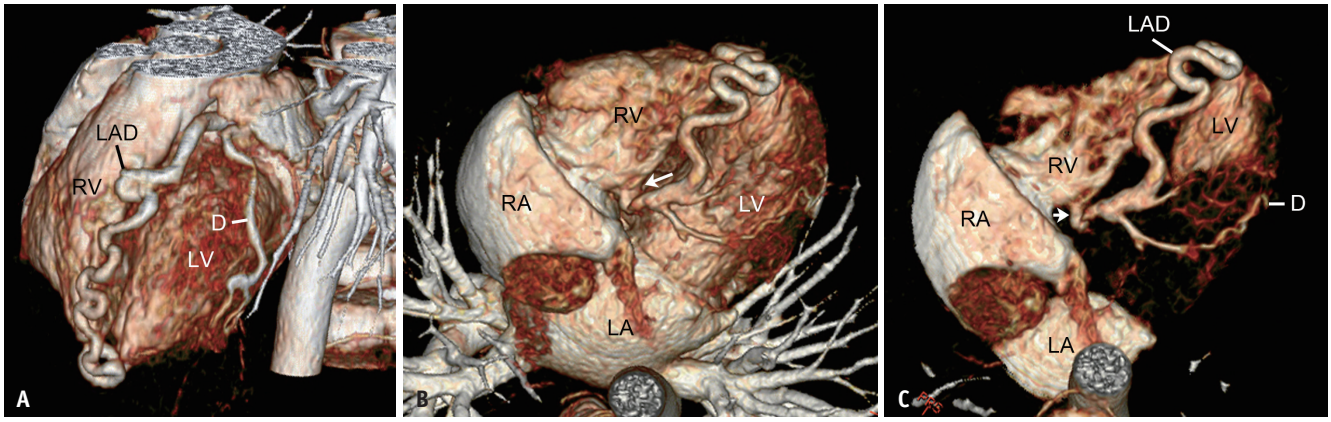

Fig. 2. Coronary cameral fistula between the LAD artery and the RV in a 5-year-old boy.
A. Oblique lateral volume-rendered CT image showing the diffusely dilated LAD artery and a D branch in a 5-year-old boy. **B, C.** Inferior volume-rendered CT images with a full slab **(B)** and with a thinner slab **(C)** demonstrating the single drainage site (arrows) of the fistula into the inferomedial basal portion of the RV. D = diagonal, LA = left atrium, LAD = left anterior descending, LV = left ventricle, RA = right atrium, RV = right ventricle

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According to the drainage site, the shunt flow may be left to right or left to left, and may even be right to left in cases with hypertensive right ventricles. When the shunt is large enough to decrease perfusion distal to the fistula, a coronary steal phenomenon may occur. Owing to this hemodynamic effect of coronary artery fistulas, various complications, including myocardial ischemia, heart failure, arrhythmia, and infective endocarditis, have been described in adults [7].

Diagnostic Imaging Modalities

Transthoracic echocardiography may show a large coronary artery fistula of typically greater than 3 mm in diameter [16]. However, its diagnostic accuracy for identifying coronary artery fistula is low, and was only 19.1% in a study of 72 adult patients with coronary-to-pulmonary artery fistula [13]. Reports on the diagnostic accuracy of transthoracic echocardiography for identifying coronary artery fistula in children are lacking. Although the acoustic window of transthoracic echocardiography is less limited in children compared to adults, its diagnostic accuracy is expected to remain insufficient to be used as a confirmatory imaging test for coronary artery fistula in children. Therefore, additional imaging methods, including catheter coronary angiography, coronary CT angiography, and coronary MR angiography, are required to make accurate diagnosis of coronary artery fistula.

The reported diagnostic accuracy of catheter coronary angiography is in the range of 35.0%–50.0% [17,18]. Due to procedure-related complications, radiation exposure, and the use of iodinated contrast agents, diagnostic catheter coronary angiography should not be primarily used for coronary artery fistulas, especially in young children.

Coronary MR angiography does not involve ionizing radiation, which may be considered a merit in pediatric imaging. However, its diagnostic accuracy in children is relatively low, mainly due to its low spatial resolution [19]. Lengthy examination time, commonly requiring deep sedation or general anesthesia, is another limitation of pediatric coronary MR angiography [20]. Consequently, cardiac MRI has been rarely used for the diagnosis of coronary artery fistula [21].

Coronary CT angiography is currently considered the imaging modality of choice for evaluating coronary artery fistula in adults [4,6-9,12-14]. Coronary artery visibility on electrocardiography-synchronized CT is also significantly improved in children [22-24]; therefore, cardiac CT has been useful for evaluating coronary artery anomalies such as coronary artery fistula in children [5,15,20,25-27]. Three-dimensional CT imaging with a thinner slab over the area of interest is especially useful for the treatment planning of

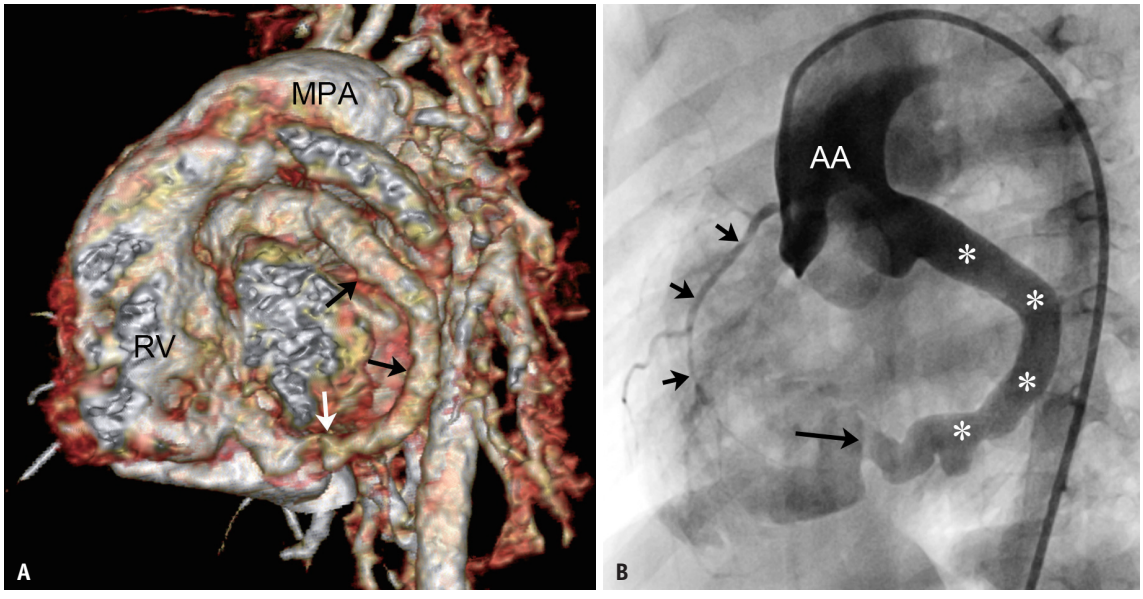

Fig. 3. Coronary cameral fistula between the left circumflex artery and the RV in a 1-month-old boy.
A. Oblique lateral volume-rendered CT image with frontal cut showing the diffusely dilated left circumflex artery (arrows) anomalously connecting to the inferomedial basal portion of the RV in a 1-month-old boy. **B.** Oblique lateral catheter ascending aortographic image showing the diffusely dilated left circumflex artery (asterisks) draining into the inferomedial basal portion of the RV (long arrow). A normal right coronary artery (short arrows) was noted. AA = ascending aorta, MPA = main pulmonary artery, RV = right ventricle

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Korean Journal of Radiology
KJR

coronary artery fistulas. However, coronary artery fistulas with mildly dilated distal coronary artery segments and small drainage sites, particularly seen in young children, may be missed given the significantly low visibility of distal coronary artery segments on cardiac CT [28]. In this case, ascending aortography or selective catheter coronary angiography may be helpful in making the correct diagnosis.

Coronary Cameral Fistula

Coronary cameral fistula is the most common type of coronary artery fistula in children, accounting for 75.0%–100.0% of cases [5,15]. Coronary cameral fistula in children most commonly originates from the right coronary artery, followed by the left coronary artery (Figs. 1-5) [5,15]. In this type of fistula, the coronary arterial origin is usually single and rarely multiple [5]. When the fistula occurs in association with pulmonary atresia and an intact ventricular septum, the origin is frequently multiple. The most common draining site of coronary cameral fistula in children is the right ventricle, followed by the left ventricle [15] (Figs. 1-5), and less frequently, the right or left atrium (Fig. 6). Coronary cameral fistula can be easily detected on imaging studies because the affected coronary artery is usually large.

Coronary-to-Pulmonary Artery Fistula

As previously mentioned, coronary-to-pulmonary artery

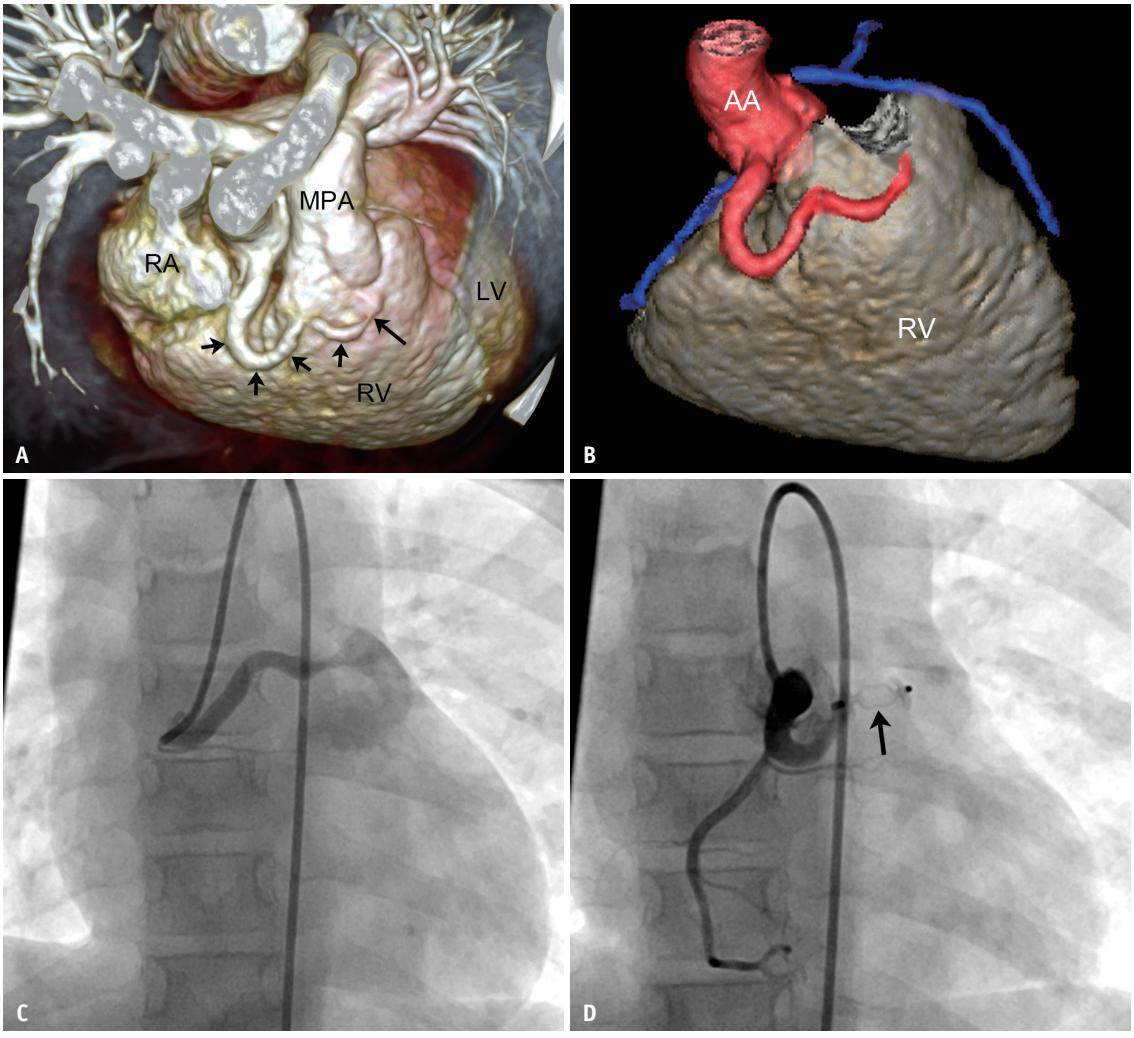

Fig. 4. Coronary cameral fistula between the conal branch of the right coronary artery and the RV.
A. Initial superior volume-rendered CT image showing the dilated conal branch (short arrows) draining into the outflow tract of the RV (long arrow) in a 9-month-old girl. **B.** Color-coded volume-rendered CT image demonstrating the coronary cameral fistula and the AA in red, and the normal right and left coronary arteries in blue. **C, D.** Frontal selective catheter right coronary arteriographic images performed 5 years later confirming the coronary cameral fistula **(C)**, and an occluding device (arrow) was placed in the distal part of the fistula **(D)**. AA = ascending aorta, LV = left ventricle, MPA = main pulmonary artery, RA = right atrium, RV = right ventricle

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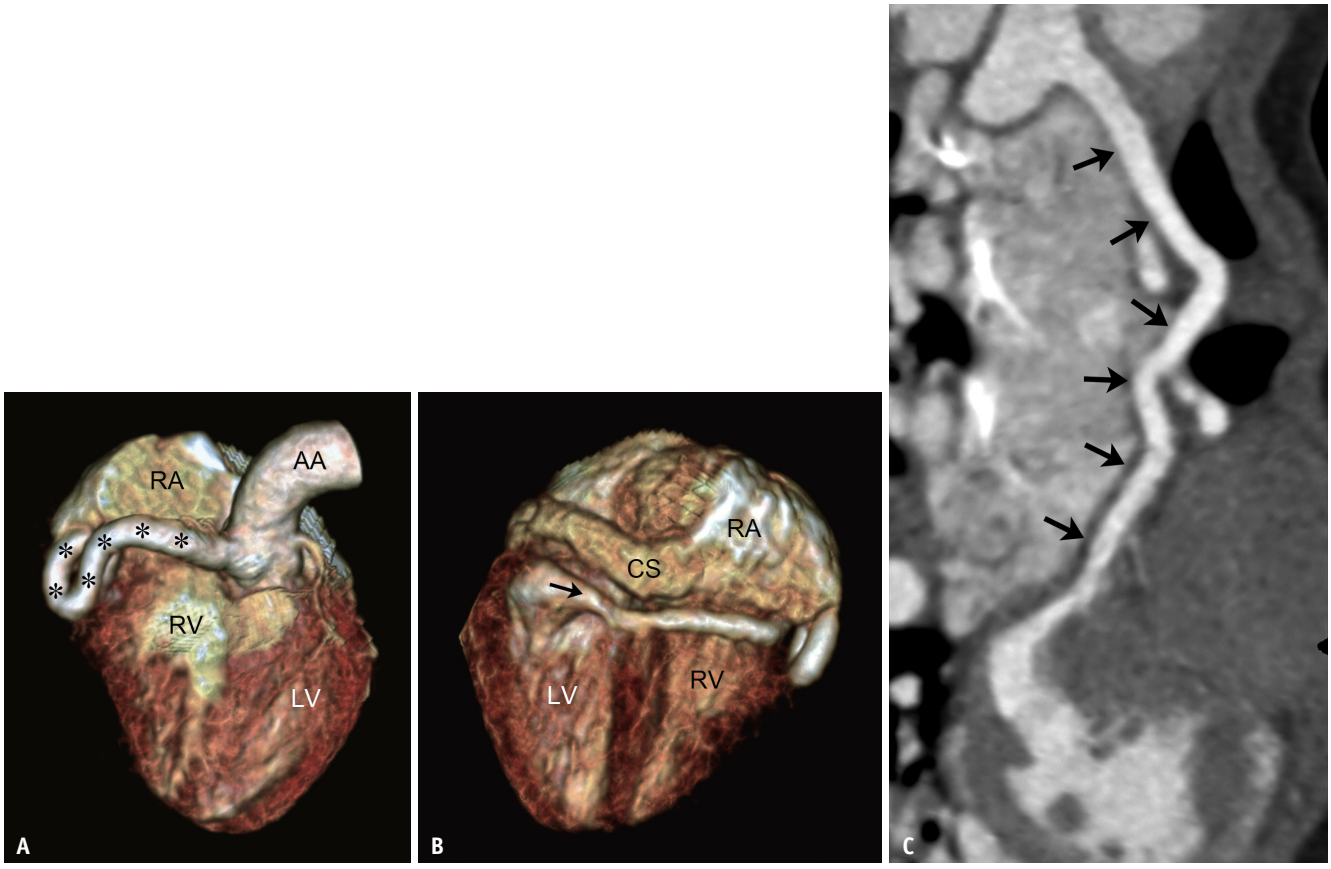

Fig. 5. Coronary cameral fistula between the right coronary artery and the LV in an 8-month-old girl.
A. Superior volume-rendered CT image showing the diffusely dilated right coronary artery (asterisks) in an 8-month-old girl. **B.** Inferior volume-rendered CT image revealing the abnormal connection (arrow) between the right coronary artery and the LV. **C.** Curved planar reformatted CT image demonstrating the entire course (arrows) of the fistula. AA = ascending aorta, CS = coronary sinus, LV = left ventricle, RA = right atrium, RV = right ventricle

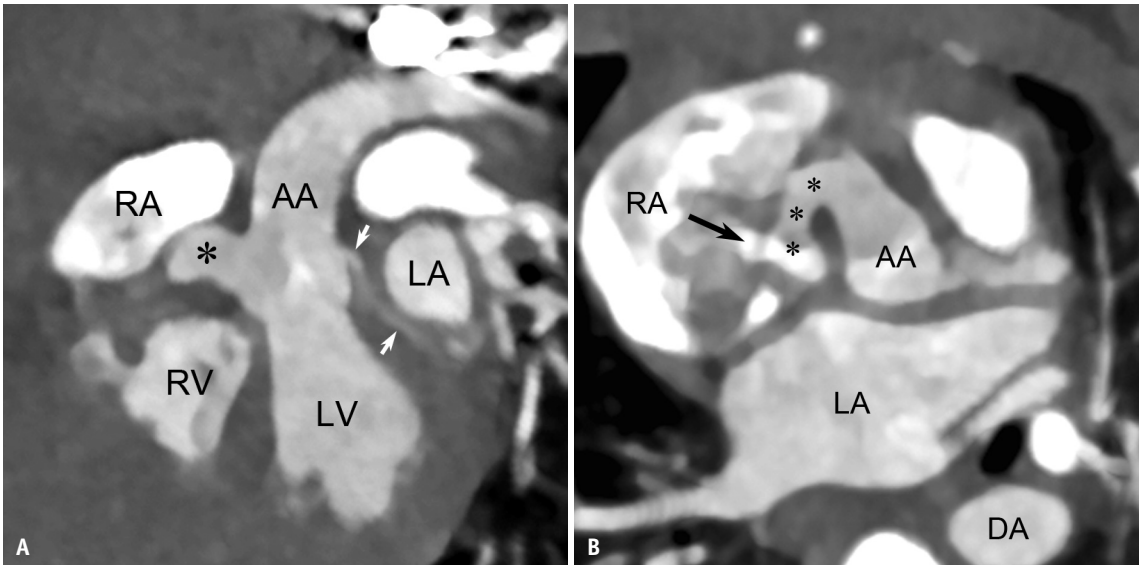

Fig. 6. Coronary cameral fistula between the right coronary artery and the RA in an 11-day-old boy.
A. Oblique coronal CT image showing the severely dilated origin and proximal portion of the right coronary artery (asterisk) in an 11-day-old boy. The left coronary artery (arrows) was normal in size. **B.** Oblique axial CT image revealing the fistulous connection (arrow) between the dilated proximal right coronary artery (asterisks) and the RA. AA = ascending aorta, DA = descending aorta, LA = left atrium, LV = left ventricle, RA = right atrium, RV = right ventricle

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Korean Journal of Radiology
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fistula is predominantly found in adults and rarely in children. In contrast to coronary cameral fistula, this fistula type most commonly arises from the proximal left anterior descending artery (45.3%–93.1%) and the proximal right coronary artery (24.5%–76.4%), and multiple origins are fairly common (30.2%–69.4%) [5,8,12,13,29]. Coronary-to-pulmonary-artery fistulas typically course anteriorly to the main pulmonary artery and drain into the anterolateral aspect, and often form a vascular network with multiple origins. Coronary arteries supplying the central pulmonary artery, as one of the major aortopulmonary collateral arteries, have been identified with ascending aortography or selective catheter coronary angiography in 10.0% of pediatric patients with pulmonary atresia and ventricular septal defect [30]. However, the tiny connection between the coronary artery and the central pulmonary artery may be easily missed on non-selective catheter angiography and other imaging modalities, including echocardiography, CT, and MRI [13].

Coronary Artery-to-Coronary Sinus (or Any Other Cardiac Vein) Fistula

Coronary artery fistulas terminating in the coronary venous system are relatively rare, accounting for 7.0% of cases [7]. The fistula may terminate in the coronary sinus (Fig. 7) or in its tributary cardiac veins (Fig. 8). Coronary artery-to-coronary sinus fistulas frequently present with congestive heart failure [31].

Ventriculo-Coronary Arterial Connections in Pulmonary Atresia with an Intact Ventricular Septum

Ventriculo-coronary arterial communications in pulmonary atresia with an intact ventricular septum are defined as abnormal vascular connections between the hypoplastic right ventricle and the coronary artery. Ventriculo-coronary arterial communications are present in approximately 40.0%

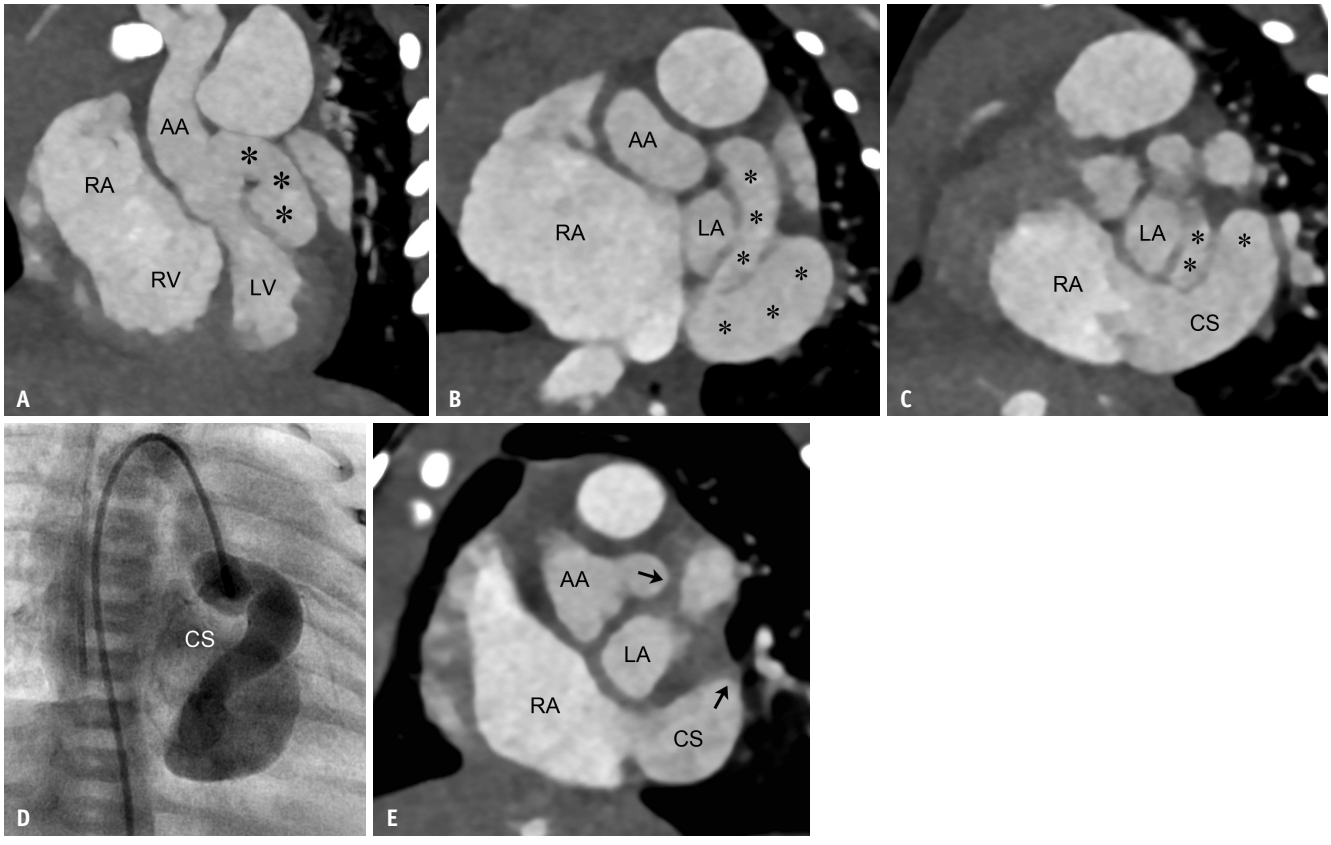

Fig. 7. Coronary artery-to-coronary sinus fistula.
A-C. Initial oblique coronal CT images showing the severely dilated left circumflex artery (asterisks) anomalously draining into the dilated CS in a 3-day-old girl. **D.** Frontal selective catheter left circumflex arteriographic image performed on the same day confirming the diagnosis of coronary artery-to-coronary sinus fistula. **E.** Follow-up oblique coronal CT image performed one month after surgical ligation of the fistula showing stumps (arrows) of the left circumflex artery and the CS. AA = ascending aorta, CS = coronary sinus, LA = left atrium, LV = left ventricle, RA = right atrium, RV = right ventricle

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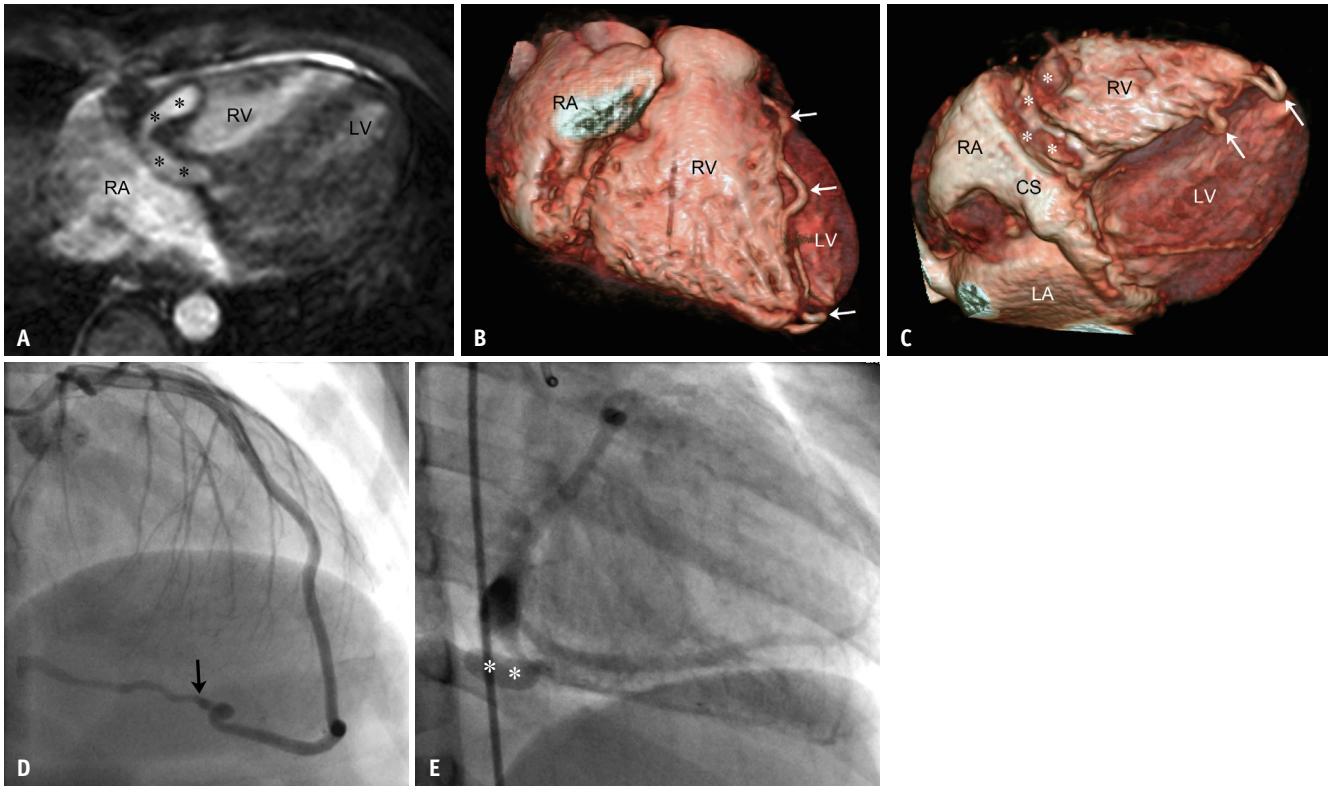

Fig. 8. Coronary artery-to-middle cardiac vein fistula.
A. Axial coronary MR angiographic image showing a fusiform enlarged vascular space (asterisks) at the basal portion of the RV in a 2-year-old boy. **B, C.** Cardiac volume-rendered CT images performed 3 years later showing the mildly dilated left anterior descending artery (arrows). However, the enlarged vascular space (asterisks) detected on MRI was barely seen. **D, E.** Frontal selective catheter left coronary arteriographic images performed 4 years later demonstrating the abnormal connection (arrow) between the left anterior descending artery and the middle cardiac vein, delayed opacification of the enlarged vascular space (asterisks), and the cardiac veins. CS = coronary sinus, LA = left atrium, LV = left ventricle, RA = right atrium, RV = right ventricle

of cases [32]. Such communications typically occur in cases with severe hypoplasia and hypertrophy of the right ventricle [32]. Right ventricle-dependent coronary arterial circulations may cause right ventricular steal phenomenon if the affected coronary arteries are patent, and may cause myocardial ischemia or infarction when coronary artery obstruction coexists. For the imaging diagnosis of a right ventricle-dependent coronary arterial circulation, there should be angiographic evidence of myocardial perfusion through the fistulous communication. Right ventricle-dependent coronary arterial circulation occurs in slightly over half of the cases with ventriculo-coronary arterial communications, and is considered a risk factor for poor outcomes in patients undergoing an attempted biventricular repair, in which the right ventricle is decompressed and the coronary arterial pressure is subsequently dropped [32]. Therefore, Fontan operation without cardiopulmonary bypass is now preferred to avoid adverse coronary arterial events, as right ventricular unloading during cardiopulmonary bypass reduces coronary perfusion pressure in right ventricle-dependent areas, and results in myocardial ischemia [32]. Ventriculo-coronary arterial connections have been traditionally observed with cardiac catheter angiography [32], but recently, such abnormal fistulous connections can be delineated with coronary CT angiography (Figs. 9, 10) [5].

Treatment

Coronary artery fistulas should be treated if the fistula is large with substantial shunt flow that may lead to myocardial ischemia, ventricular dysfunction, and congestive heart failure [7,15,33]. Surgical ligation was solely performed before the development of transcatheter closure techniques, and is now used for fistulas not amenable to interventional treatment (Fig. 7). Since its introduction in the early 1980s, transcatheter closure of the fistula has been widely used as an effective and safe treatment for coronary artery fistulas (Fig. 10) [33].

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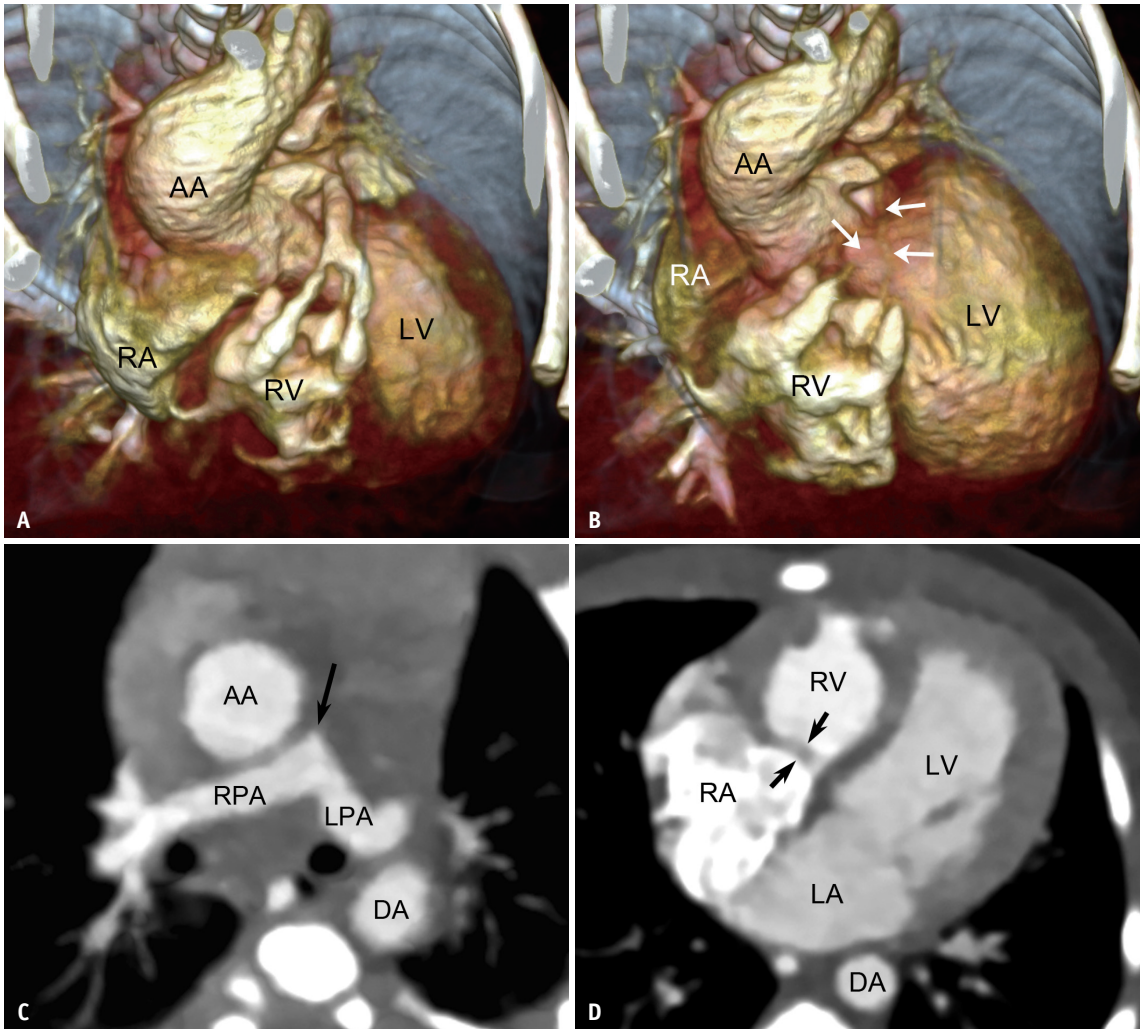

Fig. 9. Ventriculo-coronary arterial connections in a 1-day-old girl with pulmonary atresia, an intact ventricular septum, and hypoplastic tricuspid valve.
A, B. Oblique superior volume-rendered cardiac CT images obtained at end-systolic **(A)** and end-diastolic **(B)** phases showing the severely dilated left anterior descending artery and the mildly dilated right coronary artery connected to the hypoplastic RV at multiple sites in a 1-day-old girl. Notably, diastolic flow to the left anterior descending artery was significantly compromised (arrows), which may contribute to myocardial ischemia. **C.** Oblique axial CT image demonstrating pulmonary atresia (arrow) with confluent central branch pulmonary arteries. **D.** Four-chamber CT image showing the hypoplastic tricuspid valve (arrows), the hypoplastic RV, and the intact ventricular septum. AA = ascending aorta, DA = descending aorta, LA = left atrium, LPA = left pulmonary artery, LV = left ventricle, RA = right atrium, RPA = right pulmonary artery, RV = right ventricle

Contraindication to percutaneous transcatheter closure include fistulas draining close to the atrioventricular annulus, fistulas with origins at normal coronary artery branches in proximity to the occlusion site, extreme tortuosity or very small patient size making the procedure difficult, a wide draining site increasing the risk of coil migration, as well as multiple communications and drainage sites [7,15]. Coronary artery-to-coronary sinus fistula increases the risk for post-treatment adverse events such as coronary artery thrombosis and myocardial infarction [34].

CONCLUSION

In this article, the imaging findings of coronary artery fistula in children have been described. Coronary cameral fistula is the most common type of coronary artery fistula in children, and can be accurately diagnosed with CT. However, small or distal parts of other coronary artery fistula types in children may not be clearly depicted on CT, and selective catheter angiography may be necessary to characterize the whole angioarchitecture of such coronary artery fistulas. The detailed angioarchitecture delineated on coronary CT

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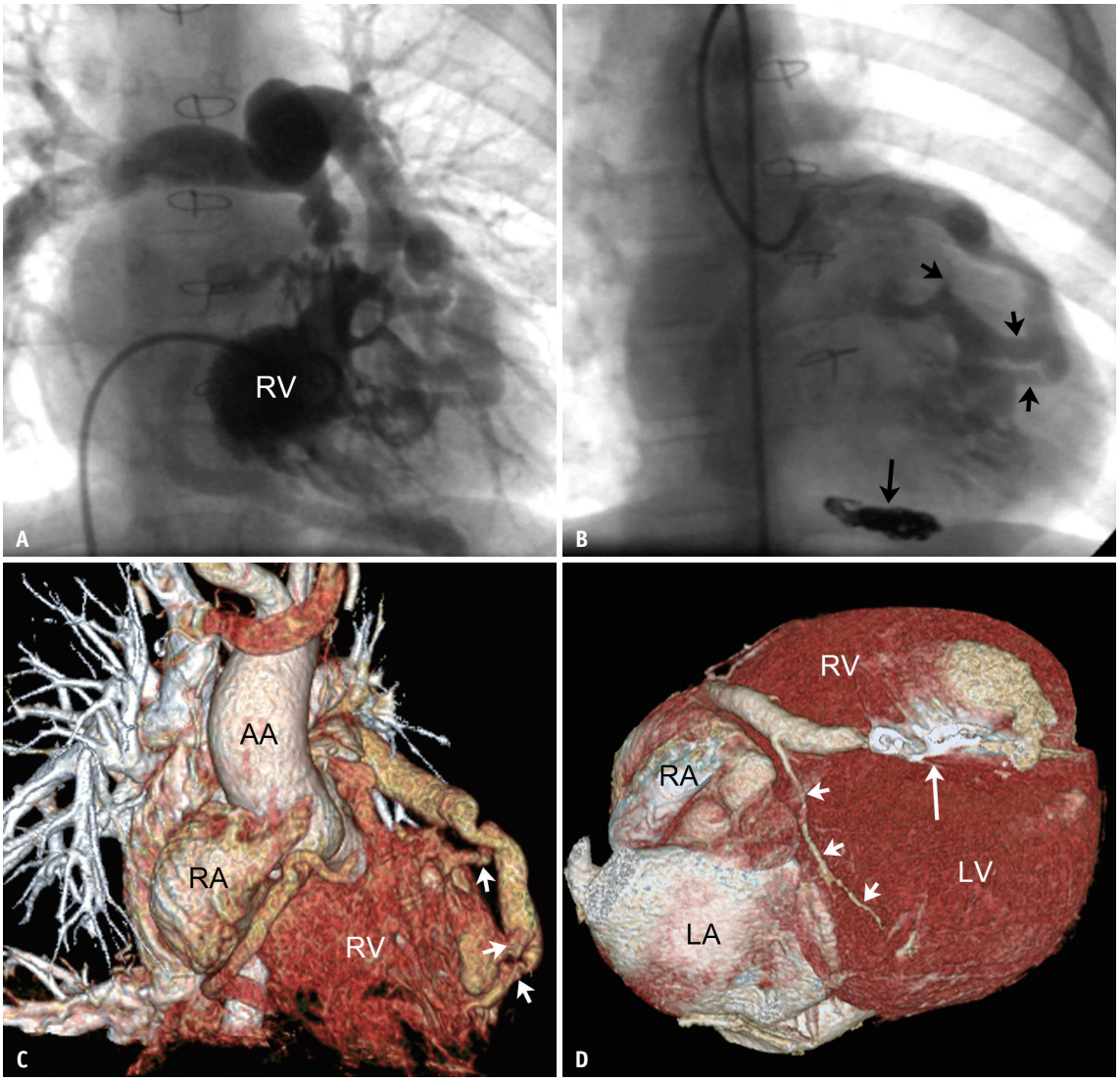

Fig. 10. Ventriculo-coronary arterial connections in pulmonary atresia and an intact ventricular septum.
A. Initial frontal catheter right ventriculographic image showing multiple abnormal connections between the hypoplastic RV and the dilated coronary arteries in a 5-year-old boy. **B.** Follow-up frontal selective catheter left coronary arteriographic image obtained after coil embolization (long arrow) of the fistulous connection between the hypoplastic RV and the right coronary artery 2 years later showing residual connections (short arrows) between the hypoplastic RV and the dilated left anterior descending artery. **C, D.** Frontal **(C)** and inferior **(D)** volume-rendered cardiac CT images performed at the age of 24 years after extracardiac conduit Fontan operation demonstrating residual connections (arrows in **C**) between the hypoplastic RV and the dilated left anterior descending artery. The right coronary artery remained dilated after coil embolization (long arrow in **D**), suggesting residual shunt flow between the RV and the right coronary artery. A normal posterolateral branch (short arrows in **D**) of the right coronary artery was noted. AA = ascending aorta, LA = left atrium, LV = left ventricle, RA = right atrium, RV = right ventricle

angiography is not only useful for differentiating coronary artery fistulas from other abnormalities manifested as dilated coronary arteries, including anomalous coronary artery origin from the pulmonary artery and Kawasaki disease, but is also crucial for treatment planning and post-treatment evaluation.

Conflicts of Interest

The author has no potential conflicts of interest to disclose.

ORCID iD

Hyun Woo Goo
https://orcid.org/0000-0001-6861-5958

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