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Use of DSA in Diagnosing and Treating Children Complex Coronary Artery Fistula: A Report of Five Cases

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Abstract

Objective: To examine the appearance of children complex coronary artery fistula using digital subtraction angiography (DSA) and to conclude whether DSA provides comprehensive information in interventional treatment.

Materials and Methods: Five children with complex coronary artery fistula were evaluated by DSA and ultrasonography (US) between 2008 and 2013. Preoperative US was performed on five children at 1, 11, 4, 12, and 5 years old following DSA evaluations that demonstrated complex coronary artery fistula. The preoperative US and DSA findings were compared with the physical examination, then decided to perform interventional treatment of three cases.

Results: In all five cases, two cases were left coronary artery to right ventricle fistulas, two cases were left coronary artery to right atrium fistulas, the other one case was right coronary artery to right atrium fistula, three cases of successful interventional treatment.

Conclusions: DSA is effective in the assessment of children complex coronary artery fistula, and may Guide interventional procedures. US may provide a useful adjunction in assessing children complex coronary artery fistula.

Keywords: Digital subtraction angiography; Ultrasonography; Children; Coronary artery fistula

Introduction

Coronary artery fistula (CAF) is a rare congenital cardiovascular malformations, the rate was 0.25% to 0.40% in congenital heart disease [1]. CAF is the presence of abnormal blood flow path between coronary artery and cardiac chambers or thirds, or other blood vessels, children are more common than adults. CAF was firstly reported by Krause in 1865, 60% to 78% of independent malformations, and other associated with atrial septal defect (ASD), patent ductus arteriosus (PDA), ventricular septal defect (VSD), etc.. US is the primary imaging modality for evaluating CAF [2]. However, since US provides limited anatomical information, DSA has provided a useful complementary tool to assess anomalies in children in recent years, coronary angiography is the "gold standard" for the diagnosis of coronary artery fistula [3].

Here, we report the findings from examinations of six children with complex coronary artery fistulas. Two children had left coronary artery to right ventricle fistula, one children had left coronary artery to right atrium fistula, the other two had right coronary artery to right atrium fistulas. Both DSA and US were used to demonstrate the CAF. This report demonstrates the importance of DSA for successful diagnosis and treatment.

Materials and Methods

This study presents five consecutive cases diagnosed for coronary artery fistula, who were referred for DSA and US evaluations between July 2008 and August 2012. Before the examination, all the patients provided their informed consent. In all the cases, the diagnosis was confirmed by combined findings of postoperative physical examinations, serial ultrasound examinations, or pathology.

Coronary angiography was performed with a 850 mA digital subtraction angiography (Neusoft, NSX6000). General anesthesia, the right femoral artery puncture, indwelling arterial sheath, routine cardiac catheterization, angiography and interventional cardiovascular treatment. Aortic root angiography and selective left and right coronary angiography, observing coronary anatomy, coronary artery fistula origin, course and drainage site; presence or absence of coronary artery aneurysm, aneurysm formation. After repeated coronary angiography, plugging effect was observed, while observing the patient's symptoms, signs, and ECG (Echocardiography) changes. When determining the CAF was completely blocked off, the patient no discomfort, no ischemic ECG changes and other signs, then the occluder is completely released. The cardiac ultrasounds were performed with a GE Vivid 7 ultrasound system with a 3–5 MHz curved-array transducer.

All of the DSA images were interpreted by two authors (Lv HT and Yang FB). The US results were known to the DSA imaging radiologist at the time of acquisition and at the time the DSA images were interpreted.

Results

All five children showed different and complex DSA and US findings. In each case, the DSA detected abnormal traffic vessels between coronary artery and heart chamber or large blood vessels. Echocardiography could only find coronal artery fistula abnormal blood flow but poor specificity for small fistula or shunt. In one of the cases, we detected a complex left coronary artery to right ventricle fistula, and branches of the left coronary artery tortuosity and expansion, the drain port is small (3.8 mm in diameter). The second case was also left coronary artery to right ventricle fistula, the performance of DSA was similar to the first case,

and the drain port is small (3.2 mm in diameter). In all five cases, two cases were left coronary artery to right atrium fistulas, the other one case was right coronary artery to right atrium fistula, three cases of successful interventional treatment.

Case 1

A 1-year-old boy, aortic root DSA revealed a left coronary artery to right ventricle fistula with significant expansion of the left coronary artery, running tortuous(Figure 1a), distal stenosis (3.8 mm diameter). Cardiac ultrasound showed dilatation of coronary section, Doppler US imaging demonstrated the direction of blood flow (Figure 1b). By X-ray fluoroscopy-guided interventional treatment of coronary artery fistula was implemented, firstly established channels by the aorta, the left coronary artery and right ventricle fistula, then placed the guide sheath. An amplatzer occluder was placed on the lower end of the narrowest part of the left coronary artery, first opening an umbrella at the distal of the stenosis (Figure 1c), another umbrella at the proximal stenosis, fistula was blocked. Postoperative aortic root DSA showed no obvious contrast agent shunt, amplatzer umbrella being normal position (Figure 1d).

Case 2

An 11-year-old boy, aortic root DSA revealed a left coronary artery anterior descending branch to right ventricle fistula (Figure 2a). Selective left coronary artery anterior descending branch angiography showed obvious expansion of the left coronary artery, running tortuous, a small right ventricle fistula (Figure 2b). Cardiac ultrasound showed dilatation of left coronary artery anterior descending branch, a small right ventricle fistula (3.2 mm diameter) (Figure 2c). Under X-ray fluoroscopy a amplatzer occluder was tried to place on the right ventricle fistula (Figure 2d), finally, no success.

Case 3

A 4-year-old girl, aortic root DSA revealed a left circumflex coronary artery to right atrium fistula with significant expansion of the left circumflex coronary artery, running tortuous (Figure 3a), distal right atrium fistula stenosis. Cardiac ultrasound showed dilatation of left circumflex coronary artery section, Doppler US imaging demonstrated the direction of blood flow (Figure 3b). This case had no interventional treatment.

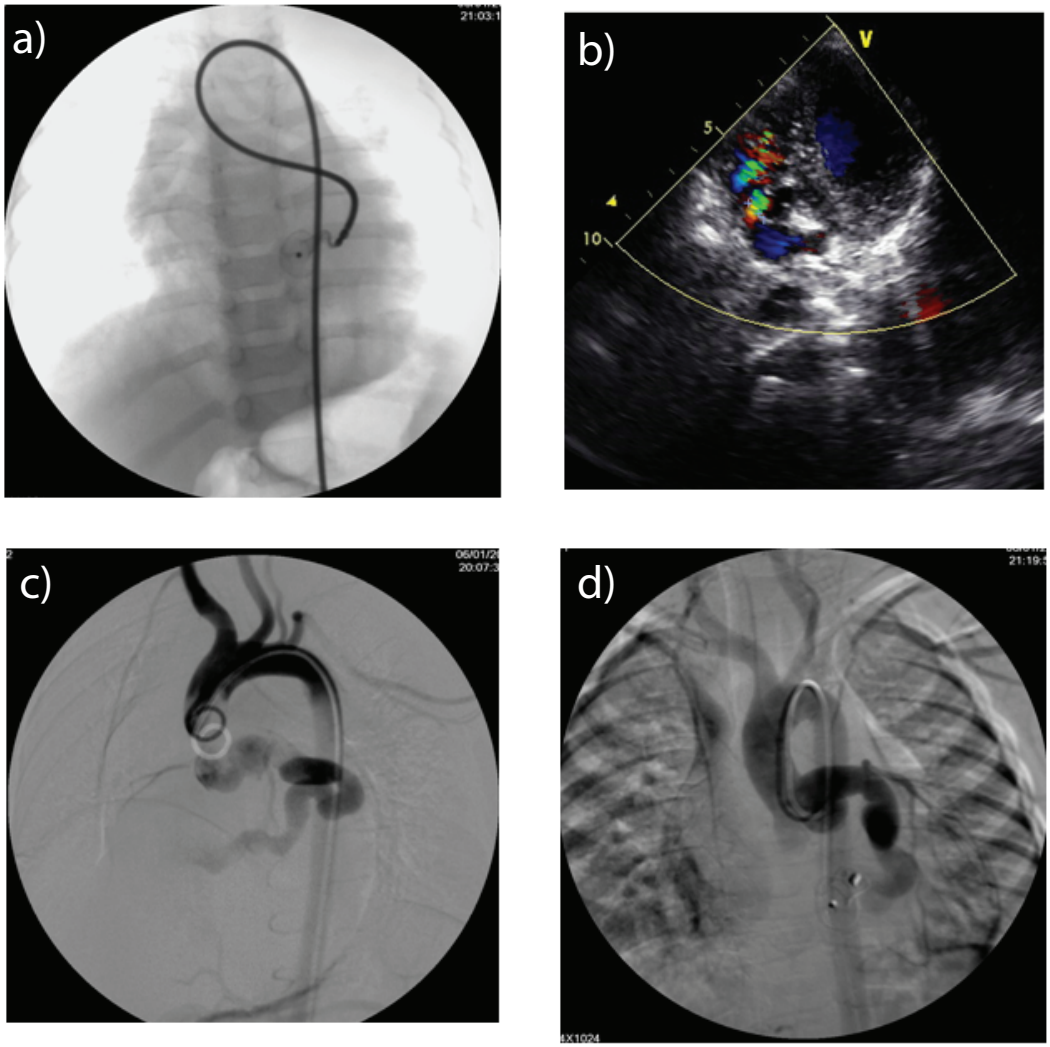

Figure 1: A 1-year-old boy with a left coronary artery to right ventricle fistula. (a) Aortic root DSA revealed expansion of the left coronary artery, running tortuous (arrows). (b) Cardiac US showed dilatation of coronary section, Doppler US demonstrated the direction of blood flow. (c) The first umbrella of amplatzer occluder was opened at the distal of the stenosis. (d) Postoperative aortic root DSA showed no obvious contrast agent shunt, amplatzer umbrella being normal position.

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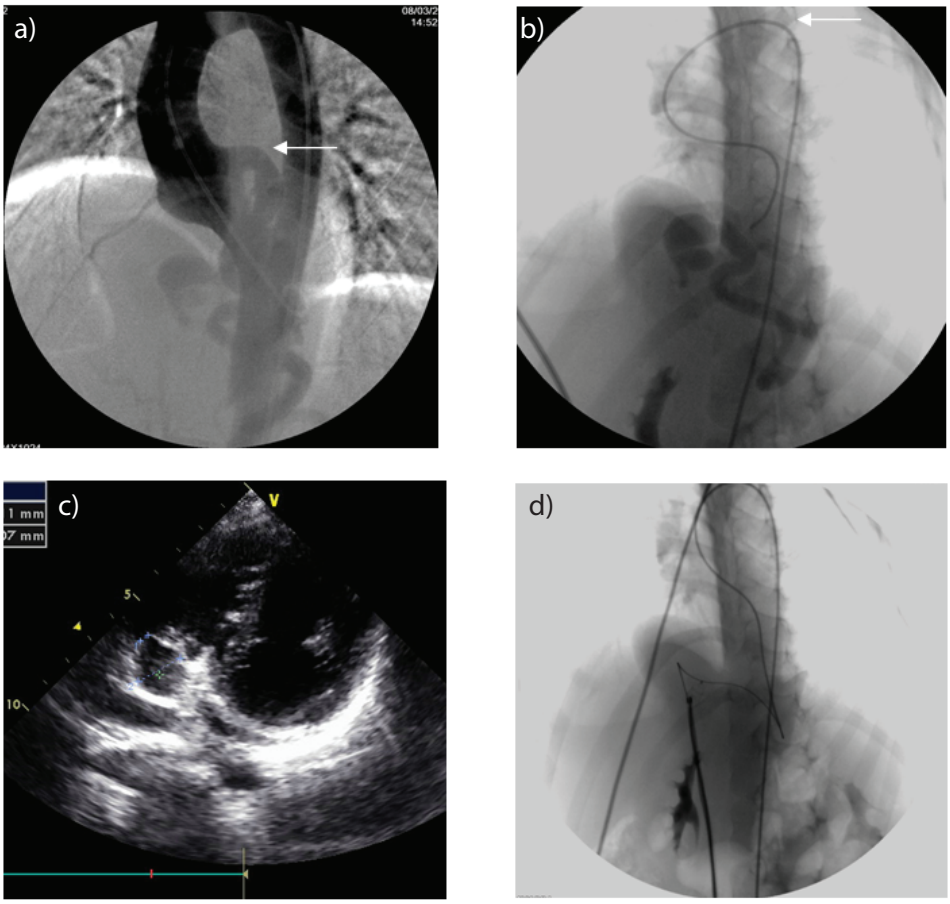

Figure 2: An 11-year-old boy with a left coronary artery to right ventricle fistula. (a) Aortic root DSA revealed expansion of the left coronary artery, running tortuous (arrows). (b) Selective left coronary artery anterior descending branch angiography showed a small right ventricle fistula. (c) Cardiac ultrasound showed dilatation of left coronary artery, a small right ventricle fistula (3.2 mm diameter). (d) Under X-ray fluoroscopy an amplatzer occluder was tried to place on the right ventricle fistula.

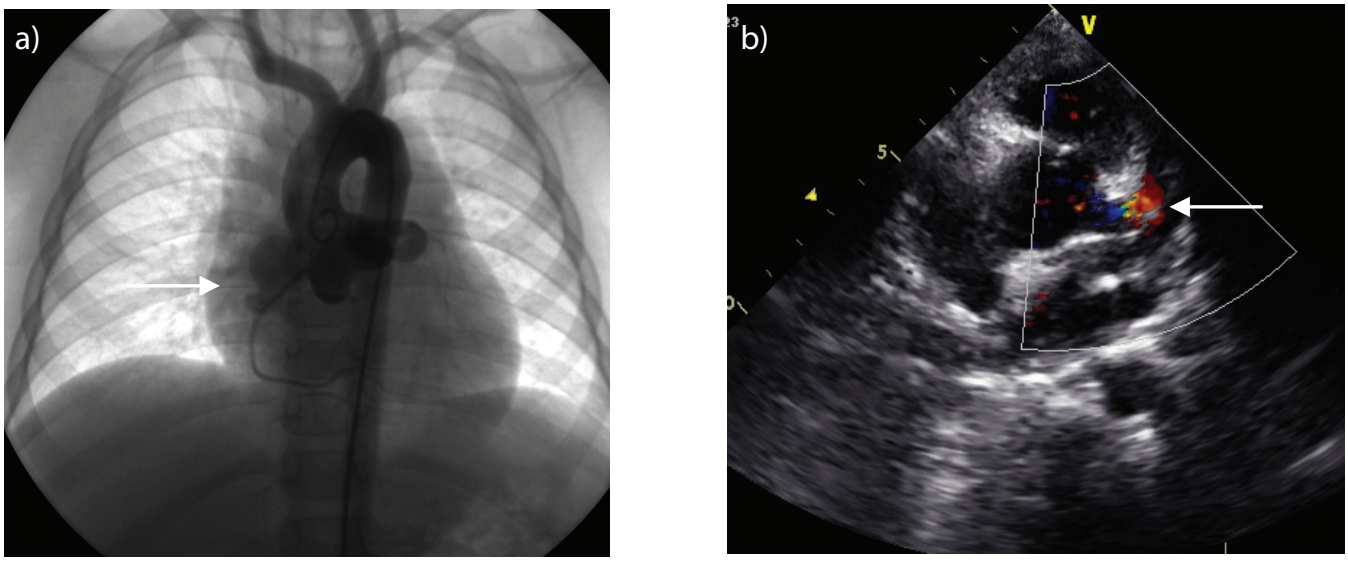

Figure 3: A 4-year-old girl with a left circumflex coronary artery to right atrium fistula. (a) Aortic root DSA revealed expansion of the left circumflex coronary artery, a right atrium fistula (arrows). (b) Cardiac US showed dilatation of coronary section, Doppler US demonstrated the direction of blood flow (arrows).

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Case 4

A 12-year-old boy, aortic root DSA revealed a left circumflex coronary artery to right atrium fistula with significant expansion of the left coronary artery (Figure 4a). Selective left coronary artery angiography showed obvious expansion of the left coronary artery, running tortuous, a small right atrium fistula (Figure 4b). By X-ray fluoroscopy-guided interventional treatment of coronary artery fistula was implemented. An amplatzer occluder was placed on the fistula, an umbrella at the right atrium, another umbrella at the expansion of left coronary artery, fistula was blocked (Figure 4c). Postoperative aortic root DSA showed no obvious contrast agent shunt, amplatzer umbrella being normal position (Figure 4d).

Case 5

A 5-year-old girl, aortic root DSA revealed a right coronary artery to right atrium fistula with significant expansion of the right coronary artery. Selective right coronary artery angiography showed obvious expansion of the right coronary artery, a small right atrium fistula (Figure 5a), fistula stenosis (3.6 mm diameter). By X-ray fluoroscopy-guided interventional treatment of coronary artery fistula was implemented, an amplatzer occluder was placed on the fistula, an umbrella at the right atrium, another umbrella at the expansion of right coronary artery, fistula was blocked. Postoperative aortic root DSA showed no obvious contrast agent shunt, amplatzer umbrella being normal position (Figure 5b).

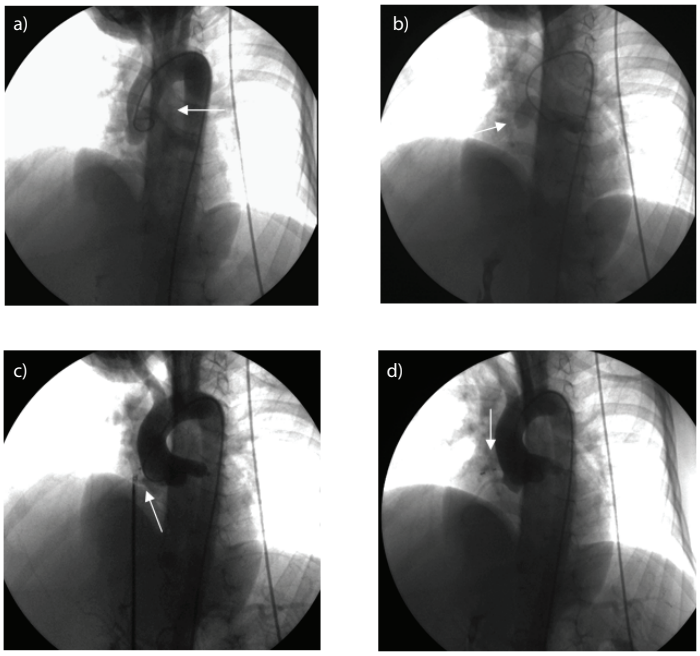

Figure 4: A 12-year-old boy with a left coronary artery to right atrium fistula. (a) Aortic root DSA revealed expansion of the left circumflex coronary artery, because of the overlap with the aorta, indicating poor (arrows). (b) Selective left circumflex coronary artery angiography showed an expansion section and right atrium fistula (arrows). (c) The umbrella of amplatzer occluder was opened at the right atrium fistula. (d) Postoperative aortic root DSA showed no obvious contrast agent shunt, Amplatzer umbrella being normal position.

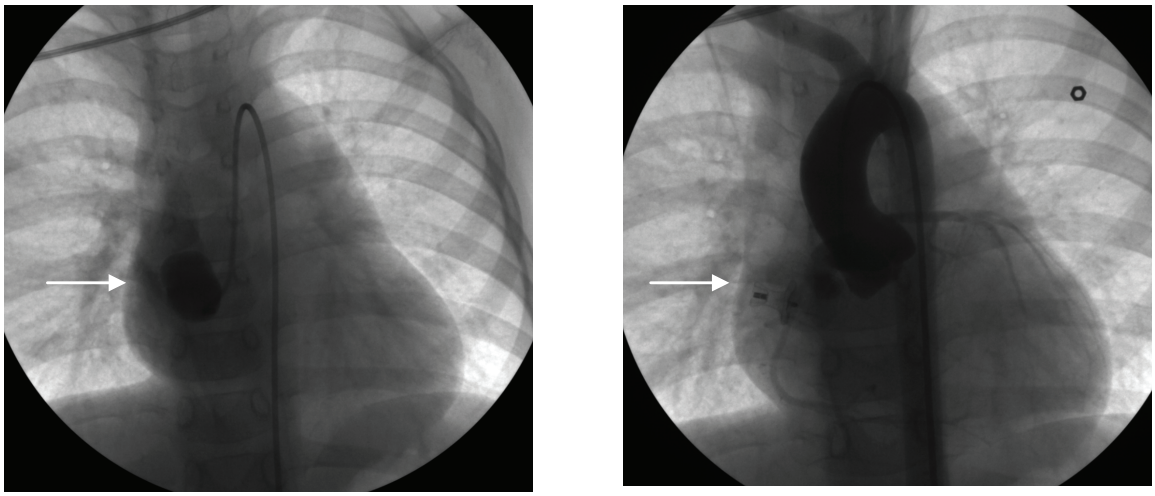

Figure 5: A 5-year-old girl with a right coronary artery to right atrium fistula. (a) Selective right coronary artery angiography showed an expansion section and right atrium fistula (arrows). (b) Postoperative aortic root DSA showed no obvious contrast agent shunt, Amplatzer umbrella being normal position (arrows).

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Discussion

Congenital coronary artery fistula is a rare congenital coronary artery disease, the main reason is due to occur during embryonic development, trabecular cardiac muscle and coronary sinus-shaped gap communicates with the cardiac development, sinus-shaped gap is compressed and gradually back into the capillaries, causing the formation of abnormal blood vessels [4]. CAF can be originated from the right coronary artery, the left coronary artery or bilateral. Clinically, coronary artery with the right atrium, right ventricle, left atrium, left ventricle or pulmonary form CAF, mainly with right heart [5]. In all five cases in this group, four cases were originated from the left coronary artery, one case was originated from the right coronary artery, all five cases were right heart fistula, and this is consistent with the literature.

CAF imaging techniques, including US, Multi-slice CT and DSA, provide critical information to diagnose fistula. Obtaining accurate imaging diagnoses of CAF remains challenging [6]. US is the primary imaging modality to evaluate CAF. Color Doppler can observe the degree of expansion of the initial segment of the right coronary artery and application flow imaging of the heart chamber and abnormal blood flow within the pulmonary artery. However, due to the expansion of the artery cannot display the full image, especially drainage to the pulmonary artery, a small fistula, shunt less prone coronary artery fistula and easy to find, easy to cause misdiagnosis. Now, with the continuous development of improved technology for the rapid development of MSCT scanning and post-processing functions, MSCT can be used as a relatively safe, fast and accurate means of checking on the diagnosis for CAF [7]. Nevertheless, US and MSCT has great limitations in the interventional treatment.

Cardiovascular DSA has provided a useful complementary tool to diagnose and treat CAF. So far, coronary angiography is considered for the "gold standard" for the diagnosis of CAF [8]. Cardiovascular DSA can display the entire coronary artery branches, clear fistula size, number, location, fistula diameter, and then guide treatment choices, especially in the presence of fistula blood vessels that supply the heart or less thick muscle has an advantage. Relative to the surgery, interventional treatment of coronary artery fistula catheter shorter hospital stay, lower costs, less invasive, but must be strictly controlled the indications, standardized operation, avoid surgical complications [9]. Amplatzer Duct-Occluder and coil are common occluder devices. Amplatzer Duct-Occluder are applicable in large coronary artery fistula usually, coil for a smaller coronary fistula. In this group, Amplatzer Duct-Occluder were used for the first, fourth, fifth cases. Amplatzer Duct-Occluder and coil were tried to place on the fistula respectively, but no success. Not all cases are suitable for interventional therapy, in conjunction with imaging findings, the patient's electrocardiogram and other clinical situations, we had the final decision on whether intervention or surgery.

Conclusions

Cardiovascular DSA is effective in the assessment of children complex coronary artery fistula, and may Guide interventional procedures. US can add useful informations in assessing children complex coronary artery fistula. With the development of intervention techniques, cardiovascular DSA are increasingly used in the treatment of children complex CAF.

Foundation Content

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