Antergrade approach for closure of distal and tortuous congenital coronary artery fistula
A case report

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Abstract
Rationale: Coronary artery fistulae (CAF) are uncommon heart lesions, but they are the most frequent significant hemodynamic congenital coronary anomaly. Transcatheter closure is an alternative treatment and a variety of techniques and devices were reported to use for the closure of congenital coronary fistulae.

Patient concerns: We reported a 5-year-10-month-old boy who was diagnosed as CAF at birth. Follow-up transthoracic echocardiography showed persistent CAF and progressed dilatation of the left coronary artery.

Diagnoses: Coronary angiography showed tortuous coronary vessel and CAF at the distal segment of the left circumflex artery (LCX).

Intervention: Antergrade approach with Amplatzer Vascular Plug was used due to the retrograde approach failed and showed a good result.

Outcomes: During a 3-year follow-up period, the patient showed no symptoms and demonstrated good exercise ability. Follow-up coronary angiography showed the Amplatzer vascular plug was located at an adequate position without a residual shunt from the CAF, and the diameter of the left coronary artery regressed when compared to previous angiography results.

Lessons: The antergrade approach is a feasible method for closing CAF if tortuous vessels are noted and fistulae occur at the distal portion of the coronary artery. A large randomized study or case series are needed to investigate the comparison of antergrade and retrograde method for the closure of CAF.

Abbreviations: CAF = coronary artery fistulae; JL = Juddins left; LCX = left circumflex artery; RV = right ventricle.

Keywords: antergrade approach, congenital coronary artery fistula, left circumflex artery

1. Introduction
Transcatheter closure approaches have emerged as a less invasive and effective strategy due to less morbidity, lower cost, shorter recovery time, and avoidance of thoracotomy and cardiopulmonary bypass. Antergrade or retrograde approaches of percutaneous closure of CAF had good long-term results. However, few reports discussed the comparison between antergrade and retrograde approach. We reported a case with tortuous coronary artery and CAF was at the distal segment of left circumflex artery (LCX). Via the retrograde approach, the closure device was difficultly transferred due to tortuous coronary vessel. An antergrade approach was performed and closed CAF successfully.

2. Case report
The 5-year-10-month-old boy described in this report had been diagnosed with a congenital coronary fistula of the left coronary artery in early childhood. CAF was diagnosed at birth. However, follow-up transthoracic echocardiography showed persistent CAF and progressed dilatation of the left coronary artery (Fig. 1A–C). Therefore, coronary angiography was performed.

Right heart catheterization showed a 1.2 pulmonary systemic flow ratio and increased oxygen saturation of the right ventricle (RV). Left ventricular angiography demonstrated a normal-sized left ventricle with good contraction, and the interventricular septum was intact. An aortogram revealed 3 competent aortic cusps without aortic regurgitation, but also showed a dilated left coronary artery with one fistula at the distal LCX. Neither patent ductus arteriosus nor coarctation of the aorta was noted. Selective coronary artery angiography demonstrated a normal right coronary artery, but the left main coronary artery, left...
anterior descending artery and LCX were dilated. A CAF draining from the distal LCX to the RV with aneurysm formation (diameter around 6.18–7.02 mm) was found (Fig. 2A–C).

Initially, we tried to close the fistula via LCX using the retrograde method. A Judkins left (JL) guiding catheter inserted deeply into the LCX with the support of 3 wires was ineffective due to torturous vessels and poor support (Fig. 3A). We switched to the antegrade approach and used a 0.025-inch Radifocus guidewire (Terumo; Japan) inserted via the right femoral vein to the RV and the pulmonary artery (PA). One snare catheter was used to catch the wire at the PA, and the Judkins right (JR) catheter was advanced into the fistula from the RV. One 8 mm Amplatzer Vascular Plug was used to close the coronary fistula (Fig. 3D). Only a tiny residual shunt was noted after the procedure (Fig. 3E), and no pericardial effusion developed.

During a 3-year follow-up period, the patient showed no symptoms and demonstrated good exercise ability. Follow-up coronary angiography showed the Amplatzer vascular plug was located at an adequate position without a residual shunt from the CAF, and the diameter of the left coronary artery regressed when compared to previous angiography results (Fig. 4A and B). The oxygen saturation was not elevated at the RV.

3. Discussion

CAF is an uncommon anomaly characterized by an abnormal connection between the coronary arteries and cardiac chambers or nearby vessels bypassing the usual myocardial capillary network and is detected incidentally in about 0.1% of adult patients undergoing routine coronary angiography. The incidence of CAF is around 0.44% during 15-year follow-up period in Asian population. Treatment of CAF is indicated for symptomatic patients and for those asymptomatic patients with a significant shunt or large fistulae that create a risk for future complications, such as infective endocarditis, pulmonary hypertension or heart failure.

There are 2 major therapeutic modalities for CAF, namely transcatheter device closure and surgical correction. Surgical correction is recommended for a large, symptomatic fistula presenting with angina pectoris, decompensated heart failure, complications due to significant pulmonary-systemic shunt, hemodynamic compromise, complex anatomy with multiple communications, tortuous pathways, multiple terminations, significant aneurysmal dilatation, or requirement for simultaneous distal bypass. In recent decades, transcatheter closure approaches have emerged as a less invasive and effective strategy. The advantages of the transcatheter approach include...
morbidity, lower cost, shorter recovery time, and avoidance of thoracotomy and cardiopulmonary bypass. Techniques previously described for transcatheter closure of CAF in adults include the use of various types of detachable occlusion coils, Amplatzer vascular plugs, Amplatzer umbrella devices, detachable balloons, covered stents, and histoacryl resin. Percutaneous closure of CAF has a good long-term outcome, and few patients experienced recanalization of the treated coronary fistulae.

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**Figure 3.** A CAG: Judkins left guiding catheter could not be inserted deeply into the LCX even with the support of 3 wires. 3B and 3C: CAG: A 0.025-inch Radifocus guidewire (Terumo; Japan) via the right femoral vein to the inferior vena cava and the right ventricle; 3I: JR catheter could be advanced into the fistula from the RV. 3D: CAG: One 8 mm Amplatzer Vascular Plug was used to close the coronary fistula via the antegrade method. 3E: CAG: Only a tiny residual shunt from the fistula was noted after the procedure. CAG = Coronary angiography, JR = Judkins right, LCX = left circumflex artery, RV = right ventricle.

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**Figure 4.** A and 4B: Follow-up CAG 3 years later: The Amplatzer vascular plug was located at an adequate position without a residual shunt from the fistula, and the diameter of the left coronary artery regressed when compared to the previous angiography results. CAF: coronary artery fistulae.
There are 2 types of angiographic classification: Type A-proximal type, where the proximal coronary segment is dilated to the origin of the fistula and the distal end is normal; and type B-distal type, where the coronary artery is dilated over its entire length terminating as a fistula mainly into the right side of the heart (end-artery type) and the proximal coronary segment can or might have regular branches. During coronary angiography, coronary anatomy, and location of CAF including origin and drainage sites need to be illustrated well. Sometimes, some devices such as microcatheters, and coronary guidewires were used to access CAF. Closure devices were selected by the operator based on the size and other characteristics of the CAF. Coils were primarily used in small to medium sized fistulae, whereas the Amplatzer devices were used to occlude larger fistulae. Few reports discussed which method of approach and the comparison between antegrade and retrograde approach. Seyed Ebrahim Kassaian et al stated the retrograde approach has some advantages over the antegrade approach if the fistula drains near the coronary sinus. In addition, percutaneous closure of CAF is a safe and effective treatment modality during long-term follow-up regardless of antegrade or retrograde approaches. Currently, no standard method or device was suggested for different type of CAF.

In our case, the guiding catheter could not be advanced to the CAF due to torturous vessels and poor support with a retrograde approach. Then, the antegrade method was switched. A snare technique was used to catch the wire, and the CAF could be closed with an Amplatzer vascular plug. Three years later, follow-up angiography showed no residual shunting and regressed dilatation of the left coronary artery.

4. Conclusion

The antegrade approach is a feasible method for closing CAF if torturous vessels are noted and fistulae occur at the distal portion of the coronary artery. A large randomized study or case series are needed to investigate the comparison of antegrade and retrograde method for the closure of CAF.

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