Transcatheter Closure of a Huge Congenital Coronary-Cameral Fistula With Amplatzer Occluder

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Abstract- Congenital coronary-cameral fistula is an abnormal connection between coronary arteries and a cardiac chamber. Although trans-catheter closure appears to be a safe alternative to surgical treatment but there is an imprecision about modality of choice. We describe a huge left coronary artery fistula to the right ventricle, found in an 8-year-old girl with dyspnea on exertion and chest pain. She was treated with an Amplatzer duct occluder. After procedure, echocardiography revealed, complete closure of fistula orifice and the girl remained completely asymptomatic during 48 months follow-up period. This study demonstrated that Amplatzer duct occluder might be a useful and safe armamentarium for coronary-cameral fistula treatment. Nevertheless, further experience and long-term follow-up studies are required to guarantee our results.

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Keywords: Huge coronary artery fistula; Trans catheter closure

Introduction

Congenital coronary fistula is a rare coronary artery malformation that connects any coronary arteries to either cardiac chambers or great vessels (1) bypassing the myocardial capillary bed (2-4).

The "cameral" word add monopolization of cardiac chamber destination to this concept (5). Although, principally it's a congenital anomaly , but it may be caused by cardiothoracic trauma (e.g. heart transplantation, myocardial biopsy) or chest irradiation (6).

Incidence of 0.2%-0.6% in angiographic series and 0.002 % in general population (6,7) and 14% in all congenital coronary anomalies has been reported for this problem (4,8). Ten to 30 percent of patients have another cardiac anomaly likewise (7).

Cameral fistula usually originate from right coronary artery (9), however, in a small proportion of patients communication may arise from left coronary artery. The most common terminal chambers are right ventricle (RV) (45%), right atrium (25%) and pulmonary artery (20%) respectively (7,10).

Because of the high pressure of left ventricle, a fistula can be aneurysmal or tortus and ultimately ruptured. Furthermore, right sided heart failure due to volume overload (3,7), myocardial ischemia due to steal phenomenon (11), fistula thrombosis, infectious endocarditis (2) and atrial fibrillation (7) are related complications (12).

Thus far, three treatment approaches are affordable; conservative, surgical and more recently trans catheter closure (3,7). Withal, there is an imprecision in preference of best suitable approach (11). We report a patient with a huge left coronary artery to right ventricle fistula treated successfully with an Amplatzer duct occluder (ADO).

Case Report

An 8-year-old girl was admitted to hospital with history of dyspnea on exertion and atypical chest pain. Clinical examination showed a grade III/VI continuous murmur at the right sternal border, the electrocardiogram showed left ventricular and right atrial hypertrophy.

Chest X-ray demonstrated moderate cardiomegaly

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and increased pulmonary vascular marking. Transthoracic echocardiography demonstrated dilated right atrium and left ventricle as well as a huge coronary artery fistula originating from the left coronary artery with drainage into right ventricle.

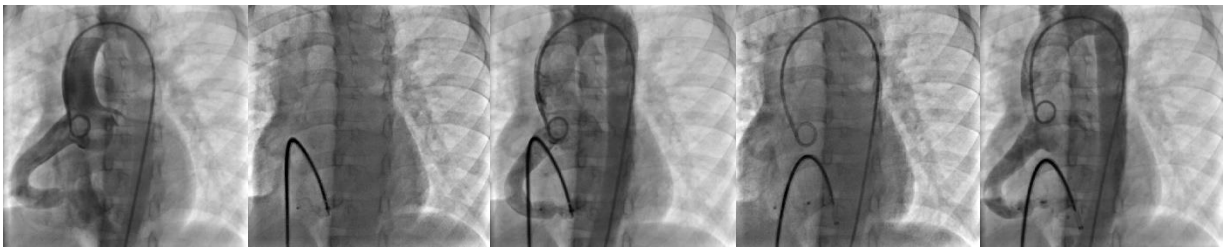
Informed written consent was obtained from parents prior to catheterization and angiography, the procedure was done under general anesthesia.

Heparin (100 IU/kg) and prophylactic antibiotic (Cefazoline 25 mg/kg) were given intravenous after the femoral venous and arterial accesses were established.

On catheterization systolic, diastolic and mean pulmonary artery pressures were 45, 15, 25 mmHg respectively. QP/QS was 3.1. Narrowest diameter of the fistula was measured 6.5 mm. Aortic root, selective right and left coronary angiogram revealed a huge left coronary artery fistula arising from proximal of the main segment and draining into the right ventricle.

Six Fr cobra catheter (Cordis® SUPER TORQUE®plus catalogue number: 533-619) was entered to the fistula from coronary artery and a 0.035-inch exchange wire (Shunmei® model no: HGWS35260) was advanced across the fistula into the left pulmonary artery. Then, wire was snared by a goose neck catheter (MEDTRONIC® no|:GN1500) and arteriovenous loop was made. A seven Fr delivery system was introduced over guiding wire loop and a patent ductus arteriosus (PDA) occluder device was advanced through the delivery system under fluoroscopic and transthoracic echocardiography. PDA occluder device was deployed into the narrowest distal site of fistula. After deployment and before release of device selective coronary angiogram was performed to assess appropriate positioning of device and residual shunt. After release of device selective left coronary angiogram showed trivial mesh leak residual shunt and good position of device. At next day echocardiography showed disappearance of residual mesh leak shunt. The device was then released from the delivery cable after confirming appropriate positioning.

The patient was put on clopidogrel and aspirin with dose of 1.5 mg/kg and 5 mg/kg for 3 and 6 months respectively to prevent thromboembolic events then maintained on aspirin for 1 year. Echocardiography was obtained in the intervals of one, six, 12, 24, 36 and 48 months after procedure that revealed fistula orifice was completely occluded. During 48 months of follow-up patient was completely asymptomatic.



1A **1B** **1C** **1D** **1E**

Figure 1. The steps of transcatheter closure of a LCA to RV fistula with huge coronary artery aneurysm (CAA).

A. Left main coronary angiography showing a dilated left coronary artery to right ventricle fistula with a huge CAA

B. Amplatzer occluder implanted at exit of coronary artery aneurysm

C. Left coronary angiography demonstrating trivial mesh leak residual shunt after device implantation and before release

D. Fluoroscopy showing suitable place of device after release

E. Repeat coronary angiography before release of device showed trivial mesh leak shunt

Discussion

Closure of small and silent coronary artery fistula in asymptomatic patients that discovered incidentally, is not recommended. Medium to large sized congenital coronary artery fistulas particularly in second decade of life (2,3,5) and that are accompanied by complications (3) should be treated using surgical or trans catheter techniques (7,11,13).

Although surgery has seldom mortality rate of one percent (11) and recurrence rate of 10 %, it has some disadvantages like median sternotomy requirement (14), cardiopulmonary bypass (5,12) in half of cases, bleeding, post pericardiotomy syndrome, myocardial infarction (7) and absence of a checking protocol for residual shunt during surgery (1).

Trans catheter device closure of coronary artery fistula (CAF) is an alternative approach to traditional surgical method. The main advantages of this method include lesser invasion, shorter hospitalization, lower cost

and faster recovery (7,13). Despite of preclusion of conceivable surgery botherations it can induce transient ischemic changes (6), unretrieved device embolization, fistula dissection, myocardial infarction due to thrombosis in the residual cul-de-sac of the occluded fistula (12), and transient atrial arrhythmia (1,3,5,7,15,16).

First successful trans catheter closure of CAF was performed by Reidy *et al*., (17). Several devices including, stainless steel coils, controlled-release coils, controlled-release PDA coils, vascular plug, ADO, and atrial septal devices are needed for trans catheter closure of CAF to be able to achieve successful closure in the vast majority of patients (3,7,11,6). Depend on age, weight, the number, size, and tortuosity of the feeding arteries, and the location of the fistula connection and its drainage site (3,7,11), different techniques and devices are used, for example; Stainless steel coils are preferable in aneurysmal fistulas and controlled-release platinum coils in those fistulas with very tortuous vessels (11).

The ADO is a self-expandable, mushroom-shaped device made from a Nitinol wire mesh, designed to close PDA (18). ADO has several advantages including the use of single device, improved control of reposition and release of the device, a high rate of complete occlusion, articulating necks, different lengths and angles accommodation and require of a small delivery sheath (4-6 Fr) (7,18).

Gribaa R *et al*., reported successful trans catheter closure of large CAF from the left anterior descending artery to right ventricle with ADO (7). Demir S *et al.*, reported a patient with huge circumflex artery to right ventricle fistula treated successfully using duct to occlude device (12).

We used the technique of arteriovenous wire loop to place the ADO antegrade in the CAF. It has several advantages: avoiding the use of large sheaths or catheters within the proximal coronary arteries, allowing entry of larger occluder via venous system and avoiding potential damage to femoral artery. Since 10 mm in size occluders should be delivered via 7-fr catheter, a trans arterial approach can damage to femoral artery, therefore venous antegrade approach avoids this complication (11,12). In report of Mottin B *et al*., success rate for complete occlusion were 87% with Amplatzer devices (6). In recent studies with improvement in techniques, delivery systems and devices, trans catheter closure of congenital coronary-cameral fistula has been performed in suitable pediatric patients (1,19).

A ten years followed up study of An 8-year-old girl affected by a giant congenital isolated coronary-cameral fistula that was draining to RV, reported minor blood passage at the prosthesis level and enlarged RV after successful closure with the ADO (20).

The ideal anticoagulant protocol in these patients is nuclease however, intensive anticoagulation with heparin and anti-platelet-aggregation drugs can cause early recurrence of mild shunt (7).

we experienced that trans-catheter closure of CAF with arteriovenous wire loop technique accomplished a feasible and safe scheme in this case. However further experience and long-term follow-up studies are required to make definitive routine use of ADO for huge CAF.

References

Nkya DA, Sinyangwe G, Takawira FF. Transcatheter occlusion of giant congenital coronary cameral fistulae: a case series. J Med Case Rep 2019;13.307

2. Bogers A, Quaegebeur JM, Huysmans HA. Early and late results of surgical treatment of congenital coronary artery fistula. Thorax 1987;42:369-73.
3. Gonçalves ES, Moura CC, Moreira JA, Silva JA. Coronary fistula to the right atrium: A challenge for the interventional cardiologist. Rev Port Cardiol 2013;32:257-9.
4. Wang S, Wu Q, Hu S, Xu J, Sun L, Song Y, et al. Surgical treatment of 52 patients with congenital coronary artery fistulas. Chin Med J (Engl) 2001;114:752-5.
5. Mavroudis C, Backer CL, Rocchini AP, Muster AJ, Gevitz M. Coronary artery fistulas in infants and children: a surgical review and discussion of coil embolization. Ann Thorac Surg 1997;63:1235-42.
6. Mottin B, Baruteau A, Boudjemline Y, Piéchaud FJ, Godart F, Lusson JR, et al. Transcatheter closure of coronary artery fistulas in infants and children: A French multicenter study. Catheter Cardiovasc Interv 2016;87:411-8.
7. Gribaa R, Slim M, Ouali S, Neffati E, Boughzela E. Transcatheter closure of a congenital coronary artery to right ventricle fistula: a case report. J Med Case Rep 2014;8:432.
8. Pagni S, Austin EH, Abraham JS. Right coronary artery to superior vena cava fistula presenting with ‘steal’phenomenon. Interact Cardiovasc Thorac Surg 2004;3:573-4.
9. Roberts WC. Major anomalies of coronary arterial origin seen in adulthood. Am Heart J 1986;111:941-63.
10. Kambara AM, Pedra CA, Esteves CA, Cano MN, Braga SL, Souza AG, et al. Transcatheter embolization of congenital coronary arterial. Cardiol Young 1999;9:371-6.
11. Qureshi SA, Tynan M. Catheter closure of coronary artery

fistulas. J Interv Cardiol 2001;14:299-307.
12. Demir S, Güler A, Uslu ZA, Kırma C. Percutaneous treatment of huge congenital coronary-cameral fistula. Turk Kardiyol Dern Ars 2014;42:168-73.
13. Qureshi SA, Reidy JF. Arterio-venous fistulas and related conditions. In: Paediatric cardiology. Elsevier; 2010:1035-54.
14. McMahon CJ, Nihill MR, Kovalchin JP, Mullins CE, Grifka RG. Coronary artery fistula: management and intermediate-term outcome after transcatheter coil occlusion. Tex Heart Inst J 2001;28:21-5.
15. Ascoop AK, Budts W. Percutaneous closure of a congenital coronary artery fistula complicated by an acute myocardial infarction. Acta Cardiol 2004;59:67-9.
16. Kharouf R, Cao QL, Hijazi ZM. Transcatheter closure of coronary artery fistula complicated by myocardial infarction. J Invasive Cardiol 2007;19:E146-9.
17. Reidy J, Sowton E, Ross D. Transcatheter occlusion of coronary to bronchial anastomosis by detachable balloon combined with coronary angioplasty at same procedure. Br Heart J 1983;49:284-7.
18. Hakim F, Madani A, Goussous Y, Cao QL, Hijazi ZM. Transcatheter closure of a large coronary arteriovenous fistula using the new Amplatzer™ duct occluder. Cathet Cardiovasc Diagn 1998;45:155-7.
19. Li YF, Zhang ZW, Wang SS, Xie ZF, Zhang X, Li YF. Transcatheter closure of congenital coronary artery fistulas with a giant coronary artery aneurysm in children: experiences from a single center. Chin Med J (Engl) 2017;130:1919-25.
20. Sozzi FB, Hugues N, Iacuzio L, Bourlon F. Giant coronary-cameral fistula: 10 years after the percutaneous closure. Eur Heart J 2017;38:1547.