Case Report

Role of Transesophageal Echocardiography in Surgical Retrieval of Embolized Amplatzer Device and Closure of Coronary–cameral Fistula

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Abstract

Congenital coronary artery fistula is an uncommon anomaly. Transcatheter coil embolization or Amplatzer vascular plug device closure of fistula is often done in symptomatic patients with safe accessibility to the feeding coronary artery. Embolization of Amplatzer vascular plug device is rare. We report an 11-year-old male child who presented to us with increasing shortness of breath for 7 years. He had a history of Amplatzer vascular plug device closure of right coronary–cameral fistula 8 years back. Echocardiography demonstrated a dilated aneurysmal right coronary artery with turbulent jet entering into the right ventricle (RV) and device embolized into the left pulmonary artery (LPA). Cardiac catheterization eventually confirmed the diagnosis. Surgical closure of fistula and retrieval of device was done using cardiopulmonary bypass. Intraoperatively transesophageal echocardiogram helped in localizing fistula opening in the RV below the anterior leaflet of tricuspid valve, continuous monitoring to prevent further distal embolization of the device during surgical handling, and assessment of completeness of repair of the fistula and LPA following retrieval of the device.

Keywords: *Coronary–cameral fistula, transesophageal echocardiogram, vascular device embolization*

Introduction

Coronary artery fistula accounts for 0.2%–0.4% of congenital cardiac anomalies.[1] Coronary–cameral fistula (CCF) involves a sizable communication between a coronary artery and a chamber of the heart.[2] Most CCFs are small, asymptomatic and remain undetected until echocardiography or coronary arteriography is performed for an unrelated cause. They usually do not cause any complication and sometimes can resolve spontaneously. However, larger one may cause myocardial ischemia or congestive heart failure requiring closure often with transcatheter coil or Amplatzer vascular plug device. Embolization of Amplatzer vascular plug device is uncommon complication that occurs early after insertion. Embolization of device after 24 h of insertion is very rare.[3] We report a case of huge right coronary artery (RCA)-to-right ventricle (RV) fistula that required surgical retrieval of embolized Amplatzer device 8 years after transcatheter closure of CCF.

Case Report

An 11-year-old male child weighing 19 kg with presented with increasing shortness of breath for 7 years. He had history of device (10 mm × 7 mm, Amplatzer vascular plug) closure of CCF from RCA to RV 8 year back. Postdevice insertion, he had developed thrombosis of the right femoral artery that was treated conservatively with heparin infusion. Child on examination revealed pansystolic murmur at apical and tricuspid area. Chest radiography revealed slight cardiomegaly and a device located in the left pulmonary artery (LPA) [Figure 1a]. Transthoracic echocardiography showed a dilated aneurysmal RCA opening into RV with an abnormal continuous turbulent flow in the RV. The dilated RCA orifice (5.8 mm at origin) showed an increased Doppler velocity during whole cardiac cycle. An Amplatzer device was located in the LPA. Left ventricle (LV) and RV function appeared normal. There was no evidence of intracardiac thrombi. The ratio of pulmonary to systemic blood flow (Qp/Qs) was 1.8:1. Computerized tomography angiography

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revealed dilated main pulmonary artery, Amplatzer device located at 7 mm from the bifurcation in the LPA [Figure 1b], and distal pulmonary artery vessel showed normal contrast opacification. Coronary angiogram showed RCA running a tortuous course between aorta and pulmonary artery [Figure 2], and the left coronary artery has normal origin and course. He was planned for the surgical closure of fistula and retrieval of device from LPA.

In the Operating Room (OR) following institution of standard American Society of Anesthesiologists and invasive arterial blood pressure monitoring, anesthesia was induced using fentanyl and propofol. Injection vecuronium was used to facilitate endotracheal intubation. A two-dimensional (2D) transesophageal echocardiogram (TEE) (9T pediatric probe of GE vivid E9 echocardiography system, GE Medical Systems, Horten, Norway) after endotracheal intubation showed the dilated RCA [Figure 3] and opening of the fistula in the RV below the anterior tricuspid leaflet [Figure 4 and Video 1]. Visualization of device in the LPA and continuous monitoring during surgical handling was facilitated using modified upper esophageal view described for visualization of patent ductus arteriosus (PDA) [Video 2].[4] After institution of cardiopulmonary bypass (CPB) and diastolic arrest with potassium-based cardioplegia fistula was visualized and closed with the PTFE patch through right atriotomy approach [Figure 5a]. Device retrieval was done through incision on pulmonary artery [Figure 5b]. The patient was successfully weaned off from CPB with noradrenaline 0.05 μg/kg/min. Post-CPB, TEE showed no flow from the RCA to RV, no regional wall motion abnormality, and no gradient or turbulent flow across LPA. The Qp/Qs ratio decreased to 1.1:1. The patient was successfully weaned off from ventilator after 6 h of mechanical ventilation and shifted to ward on the 2nd postoperative day. He was discharged from hospital on the 4th postoperative day.

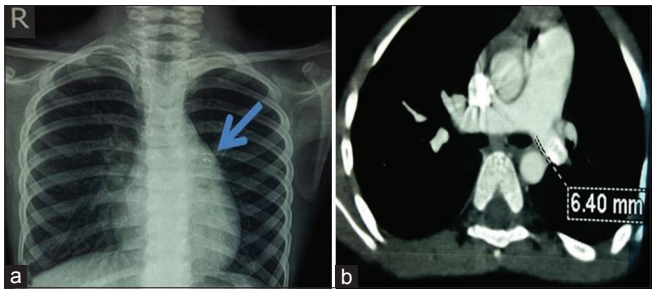

Figure 1: Chest X-ray posterior-anterior view (a) and computed tomography angiography (b) cardiac device in the left pulmonary artery

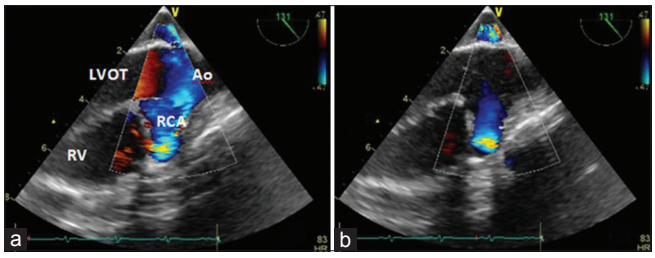

Figure 3: Echocardiography before surgery. Two-dimensional transesophageal echocardiography, aortic valve long-axis view, showing dilated right coronary artery and color Doppler flow in systole (a) and diastole (b). LVOT: Left ventricular outflow tract, Ao: Aorta, RCA: Right coronary artery, RV: Right ventricle

Discussion

Primary CCF is either isolated cardiac lesion or associated with some minor cardiac anomalies such as PDA or atrial septal defect while secondary CCF is complicated due to its association with major cardiac anomalies. CCFs may arise from RCA (55%), left coronary artery (35%), both coronary arteries (5%), or from an anomalous single coronary artery (3%). Majority of fistula (>90%) opens in the right side of the heart, primarily the RV (42.5%), right atrium (34%), pulmonary artery (15%), or coronary sinus (7%), and very rarely in the left atrium (5%) or LV (3.5%).[1,5,6]

The main pathophysiology changes due to the presence of CCF are reduced myocardial blood flow distal to the

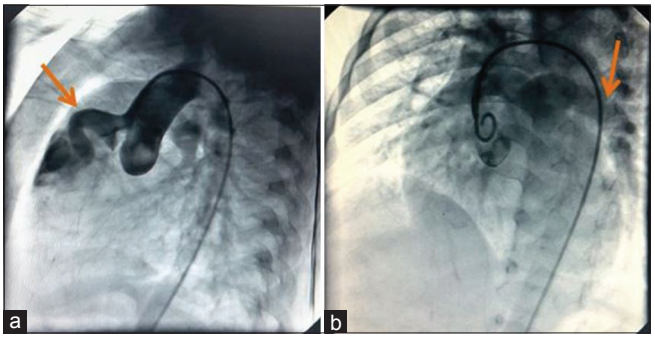

Figure 2: Preoperative angiography. (a) Aortic root angiography, anterior-posterior view, revealing an aneurysmal right coronary artery (large arrow) with a fistula shunting into the right ventricle and (b) flow in the left pulmonary artery

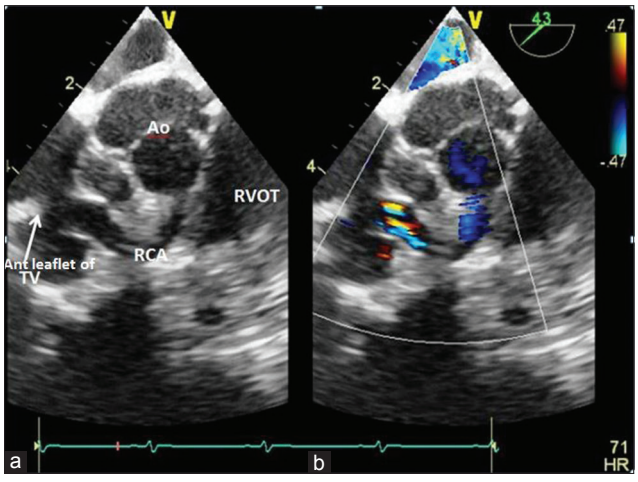

Figure 4: Echocardiography before surgery (a) Two-dimensional transesophageal echocardiography, aortic valve short-axis view, showing dilated right coronary artery opening into right ventricle below tricuspid leaflet. (b) Color Doppler echocardiography showing flow in the right coronary artery. RVOT: Right ventricular outflow tract, Ao: Aorta, RCA: Right coronary artery, TV: Tricuspid valve

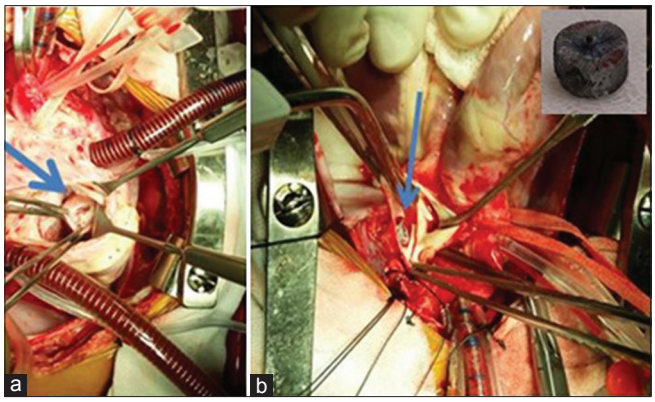

Figure 5: Tortuous course of the right coronary artery to right ventricle (a) opening just below anterior tricuspid leaflet. Single-lobe Nitinol mesh design Amplatzer vascular device (b) being removed from the left pulmonary artery

fistula. The distal myocardial blood flow depends on the fistula size, pressure gradient across fistula, and runoff from the coronary vasculature to a low-pressure cavity. Initially, coronary vessel attempt to compensate by increasing the caliber of the ostia and feeding artery. In a large fistula, the intracoronary diastolic perfusion pressure may decrease to the extent to cause myocardial ischemia, and left to right shunt causes pulmonary overperfusion eventually leading to heart failure. Liberthson *et al*.[7] found no significant difference in mean shunt size between symptomatic and asymptomatic patients with CCF. The natural history of CCF is variable.[1,4,8,9] Spontaneous closure of CCFs is very rare (1%–2%).[10,11] Patients with small fistula may remain asymptomatic throughout life. Common fistula-related complications among pediatric patients include congestive heart failure (12%), myocardial infarction (4%), bacterial endocarditis (3%), and death (6%). Other rare life-threatening complications included giant aneurysmal dilatation of the fistula, dissection, and rupture causing cardiac tamponade. Our patient presented with features of congestive heart failure which is the most frequent presentation in pediatric patients.[1,5]

The goal of treatment for symptomatic patients is closure of the fistula without hindering the native coronary artery supply. Closure of the fistula can be carried out either surgically or by transcatheter closure. Factors to consider in deciding between surgery and percutaneous closure include the size of the fistula, its location and drainage pattern, and presence of associated cardiac lesion. The reported morbidity and mortality (<3%) and incidence of recurrence (4% of patients) are low with surgical method that often require the use of CPB although successful surgical repair without the use of CPB has also been described.[12,13] A less invasive percutaneous transcatheter closure is preferred in carefully selected patients such as absence of multiple fistulae or presence of large branch vessels providing safe accessibility to the coronary artery supplying the fistula.[14] The number of devices such as coils, detachable balloons, umbrellas, polyvinyl alcohol foam, PDA device, or vascular plug has been used for this purpose.[15-18] Complications such as transient T-wave inversions or arrhythmias, coronary artery trauma or rupture, total occlusion of a coronary artery, device migration, and bacterial endocarditis after transcatheter closer of fistula have been reported. The reported incidence of embolization of occlusion device is about 17%.[3] Most of these embolization occurs early after deployment of device; however, rarely delayed embolization may occurs. Hence, it is recommended to repeat catheterization after 3–6 months of coil occlusion which was not done in our patient due to loss in follow-up.

Echocardiography is helpful delineating anatomic details and functional consequences of CCF. 2D echocardiography helps in demonstrating dilated coronary artery its course and associated enlarged or tortuous feeding coronary artery. Color Doppler helps in localizing site of drainage. Large flow is present at the origin or along the length of the fistula vessel.[19] Sometimes, cardiac catheterization and angiography may be needed to assess origin and course of the coronary artery involved, presence of any obstruction and the drainage site, and the hemodynamic significance of the fistula. Intraoperative TEE is useful to confirm the diagnosis, localization of fistula drainage site, magnitude of left to right shunt, and function of RV and LV before and after repair of fistula. In the index case, TEE helped in localizing the drainage site of fistula in the RV below the anterior tricuspid leaflet; embolized device was visualized and monitored continuously during surgical handling using modified upper esophageal view to see LPA, and finally, adequacy of repair following device retrieval and closure of fistula was assessed by the absence of shunting in the RV, absence of turbulence flow or gradient in the LPA, and regional wall motion abnormality of RV and LV.

Conclusion

Embolization of device following closure of CCF is rare. TEE plays a very crucial role during surgical closure of CCF and retrieval of embolized device.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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