

A second look at surgical repair of a distal coronary artery fistula: Stems from trunk

Sir,

The shunt in coronary cameral fistula (CCF) can be completely closed or reduced significantly either by surgical or percutaneous closure. The immediate residual ectasia or aneurysm is unavoidable in percutaneous closure. Future progression of ectasia to aneurysm, aneurysm to giant aneurysm and its related complications like endarteritis, systemic and coronary embolism, and impingement on neighboring structure are though infrequent, may need another surgical repair. Surgical resection of aneurysm of the coronary artery with multiple distal exits into cardiac chamber is preferred to device-based closure to reduce the remote residual aneurysm and its related complications.^[1] The short-term or interim follow-up results with either treatment reduces the size of dilated artery.^[2-5] Sometimes unfavorable remodeling of residual ectasia or aneurysm of conduit artery needs long-term follow-up with oral anticoagulation.^[6-8]

A 31-year-old young man presented with history of low grade fever and heaviness in the chest for last 3 months. He was previously evaluated and found to have a distal CCF of right coronary artery with multiple exits into right ventricle, the largest exit being 8×10 mm, opening just below the tricuspid valve. There was already mild dilatation of proximal conduit artery at the time of surgery in 1996. The exits and the entry points of fistula were closed surgically under cardiopulmonary bypass

without residual shunt at the age of 11 years. The case was followed-up annually for future aneurysmal dilatation of residual ectatic conduit artery. Previous blood cultures done elsewhere were negative for microorganisms. General clinical and cardiovascular system examinations were normal. The chest X-ray in lateral projection showed complete filling of retrosternal space with a non-homogeneous opacity. Twelve-lead electrocardiogram was consistent and showed Q waves in the inferior leads. Two-dimensional echocardiography confirmed the diagnosis of giant right coronary artery aneurysm with anterior extension of distal end. There was no residual shunt and the distal end was filled with organized thrombus [Figures 1 and 2]. There was no regional wall motion abnormality in the inferior wall. The total leukocyte count and neutrophil count were high with a shift to left; blood cultures were negative. The fever responded well to empirical antibiotics (ampicillin and gentamicin) given for 2 weeks, given for possible endarteritis. Sixty-four slice multidimensional computed tomography (CT) with contrast enhancement of aortic root and coronary arteries showed dominant right coronary artery, giant right coronary artery aneurysm including the adjacent of right sinus of Valsalva (RSOV) of size 3.5×2.8 cm at the ostium and 5.6×6.3 cm distally filled with thrombus. The distal half of cull-de-sac had patchy calcification. Posterior descending artery and post left ventricular branch were filling by Rentrop's grade III contralateral collaterals [Figure 3]. The aneurysm was excluded from right coronary sinus, using Dacron patch

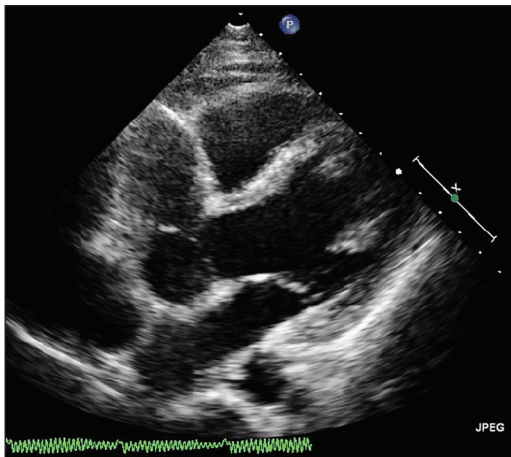


Figure 1: Two-dimensional (2D) echocardiography in modified five chamber with extreme anterior swipe of the of the probe shows aneurysmal dilatation of right coronary sinus communicating giant aneurysm right coronary artery with distal cull-de-sac containing organized thrombus just behind the sternum



Figure 2: 2D-Echocardiography in short axis view with anterior swipe of echo window is consistent with the observation made in the Figure 1

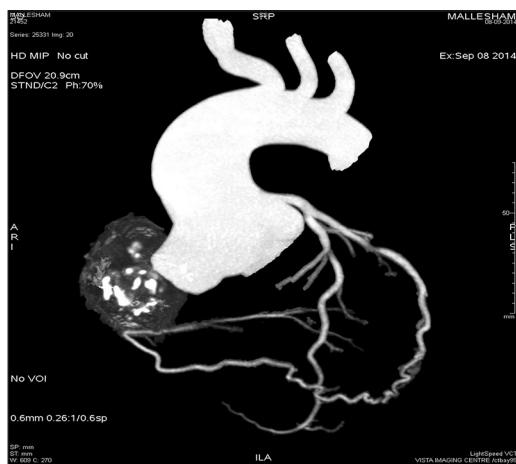


Figure 3: Light speed Volume Computed Tomography (VCT) 64-slice multidetector computed tomography (MDCT) with contrast enhancement of aortic root and coronary arteries showed dominant right coronary artery, giant right coronary artery aneurysm including the proximity of right sinus of Valsalva (RSOV) of size 3.5×2.8 cm at the ostium and 5.6×6.3 cm distally filled with thrombus. Patchy calcific segments were seen in the distal half of cull-de-sac. Posterior descending artery and post left ventricular branch were filling by Rentrop's Grade III contralateral collaterals. Other coronary arteries were normal

of size $3 \text{ cm} \times 2 \text{ cm}$. The compressed and eroded right atrial wall was repaired by using bovine pericardial patch. The distal right coronary was grafted using saphenous venous graft. The procedure under cardiopulmonary bypass was uneventful as was the postoperative recovery. The biopsy specimen of aneurysmal sac was consistent with evidence of chronic nonspecific endarteritis.

The effect of patient age on the development of coronary artery aneurysm in Kawasaki disease shows a U-shaped relation, with the nadir of age range being between 11 to 48 months. Some long term follow up of Kawasaki disease suggest Kawasaki disease also needs long term follow up for aneurysm and its related complication^[9] same as in the cases of residual dilated coronary arteries after closure of congenital CCF with high risk factors like residual ectasia or aneurysm, especially after percutaneous closure of CCF^[4,6]. The aneurysm in our case was excluded from right coronary sinus using a double-layered patch consisting of bovine pericardial patch and Dacron patch for better strength. The American College of Chest Physicians (ACCP) guideline^[10] recommendation for oral anticoagulation in children using warfarin was used for initiation and maintaining the dose warfarin in our case. The postoperative use of oral anticoagulant (OAC) has been well-emphasized in the study by Gowda *et al.*^[11] Coronary artery aneurysm associated with a coronary-cameral fistula is rare condition.^[12] It is even more uncommon to find a giant coronary aneurysm with related complication after surgical closure.^[7] The contemporary review of literature does not yield any report of such large aneurysm after closure of coronary-cameral fistula that has undergone surgical exclusion and coronary

bypass graft. The conduit artery dilatation to aneurysmal proportions, endarteritis and thrombus occluding the distal arteries needing a second look surgical repair, has not reported previously. Here it happened after 2 decades in the absence of shunt. We routinely use echocardiography in outpatient follow-up. Though selective coronary angiogram is gold standard for evaluation in this condition, three-dimensional (3D) computed tomography (CT) coronary angiogram or magnetic resonance (MR) angiogram can be done if pathology is not clear.^[13,14]

This case can be considered as a “stem from trunk”. If the trunk of a tree is nourished well, even after cutting all the branches, new branches can arise from the trunk. Therefore long-term follow-up is essential irrespective of closure techniques used for CCF when residual conduit artery is associated with ectasia.

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