Complete Closure of a Large Ventricular Septal Defect by a Septal Aneurysm

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Abstract

We describe an adult case of a large ventricular septal defect (VSD) with a secundum atrial septal defect (ASD), where the VSD was completely closed by an aneurysmal sac. Cardiac findings were incidentally detected on computed tomography pulmonary angiography (CTPA), mimicking a right ventricular mass and later confirmed by echocardiography. We discuss the available data of these aneurysms of membranous ventricular septum (AMVS), which favors the surgical treatment even though the VSD might have been obliterated by the aneurysmal sac.

Keywords: Ventricular Septal Defect; Echocardiography; Aneurysm

Introduction

An aneurysm of membranous ventricular septum is a rare, incidentally detected anomaly characterized by localized bulging of the membranous portion of the interventricular septum, often involving tissue from the septal leaflet of the tricuspid valve. The incidence was reported to be 0.17% in a longitudinal study of 30,120 adult patients.[1] In a study of 600 patients with congenital heart disease at the Royal Hospital, Muscat, none had AMVS.[2] The aneurysm formation may partially or completely seal a VSD. These aneurysms have been reported to lead to multiple complications. Management involves surgical intervention, with a more conservative approach adopted for a few asymptomatic patients who are under regular follow-up.

Case Report

A 51-year-old male presented with dizziness that started while driving his bike. The dizziness worsened with head movement and on closure of eyes. He was healthy previous to this episode and not on any medications. He denied any chest pain, palpitations, dyspnea, syncope or fever. Physical examination was unremarkable other than a grade 3/6 systolic murmur over precordium. He was acyanotic and overall in a good stable condition. Laboratory values were normal except for high D-dimer and cardiac troponin T. Electrocardiography showed normal axis with P pulmonale sign.

The patient presented with episodes of dizziness and had elevated troponin and D-dimer. A CTPA was immediately advised by the emergency physician and revealed no pulmonary embolism. Other findings were right atrial (RA) enlargement, and a filling defect in right ventricle (RV), possibly due to a thrombus or mass. Pulmonary artery was dilated, raising suspicion of pulmonary hypertension [Figure 1].

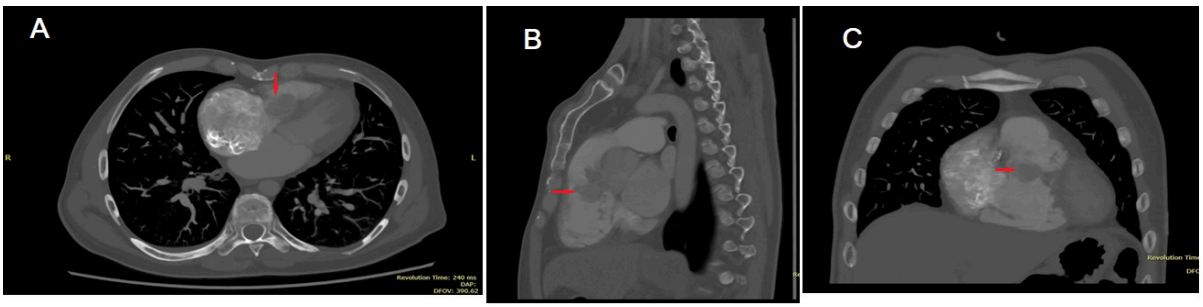

Figure 1: CT pulmonary angiography images. (A) Axial image showing dilatation of right atrium and a filling defect in RV (red arrow). (B) Sagittal image showing a lobulated aneurysmal pouch bulging into RV (red arrow). (C) Coronal image showing the same aneurysm. Image B and C show the aneurysm arising below pulmonary and aortic valves. RV, right ventricle.

Transthoracic echocardiography (TTE) revealed a perimembranous VSD, sealed by a large aneurysmal pouch. The VSD measured 12 x 15 mm, with no left-to-right shunt through the aneurysm. The aneurysmal sac measured 29 x 26 mm. Color Doppler imaging showed no communication with the RV and no flow signal was observed entering the aneurysm through the VSD [Figure 2]. There was prolapse of both mitral valve leaflets with grade 2/4 eccentric mitral regurgitation (posteriorly directed jet). No cleft was identified in the mitral leaflets. A large secundum ASD, measuring 16 × 20 mm, with a left-to-right shunt was present. The RA and RV were severely dilated, with a flattened interventricular septum (IVS). Tricuspid regurgitation was mild, with a calculated pulmonary artery systolic pressure of 48 mmHg. [Figure 3].

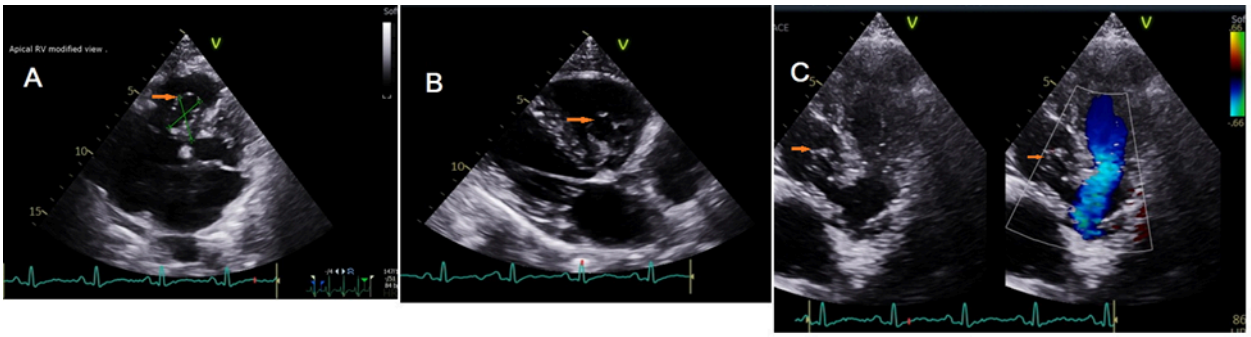

Figure 2: Transthoracic echocardiography. (A) Apical RV modified view showing an aneurysm of perimembranous ventricular septum (red arrow). (B) Modified parasternal long axis view showing perimembranous VSD and the aneurysm (red arrow). (C) Color flow imaging in apical 5-chamber view showing no flow across the VSD and into the aneurysmal pouch. All three views show the aneurysm arising below aortic annulus. ECG tracing in green; VSD, ventricular septal defect.

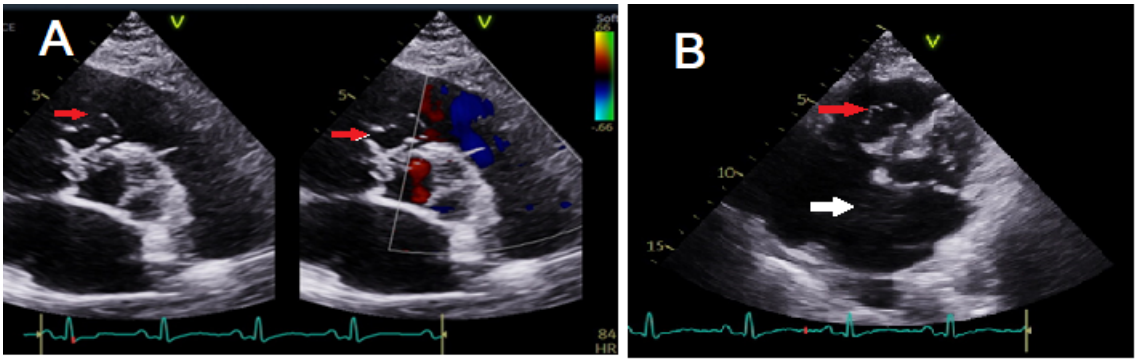

Figure 3: Transthoracic echocardiography. (A) Color flow imaging in parasternal short-axis view showing a ventricular septal aneurysm (red arrow). (B) Modified apical view showing aneurysm (red arrow) and atrial septal defect (white arrow).

Discussion

The first descriptions of an AMVS were reported from postmortem examinations.[3] Its first antemortem diagnosis was made in 1957 by Steinberg.[4] Spontaneous closure of muscular and perimembranous VSDs is frequently observed in early childhood, whereas spontaneous closure of a perimembranous VSD measuring more than 4 mm is uncommon. However, sometimes, even a large perimembranous VSD may close spontaneously by an aneurysm, most frequently originating from tissue of septal leaflet of tricuspid valve. The closure may be complete or incomplete. Other mechanisms of aneurysm formation are mycotic or bacterial endocarditis of aortic valve with involvement of membranous septum. Rarely the membranous aneurysms may be of congenital origin, originating mostly from interventricular portion of membranous septum. Infundibular defects can sometimes close by prolapse of the right aortic cusp.[5] The main differential diagnosis of AMVS is aortic sinus of valsalva aneurysm (ASVA). On TTE, the ASVA originates from above the aortic annulus, while AMVS is located below the aortic annulus. The diameter of AMVS generally varies from 1 to 3 cm. The diameters in our case were 29 x 26 mm, forming a huge sac in RV. In some patients, the aneurysmal wall may become calcified with time. In the present case, there was no evidence of calcification on CT or TTE images [Figure 1-3]. The use of multimodality imaging facilitates accurate diagnosis of this rare finding; in particular, transesophageal echocardiography provides excellent visualization of the spatial relationship between the aneurysm and intraventricular obstruction, allowing optimal preoperative planning.

Closure of a VSD by aneurysm formation may be perceived as a welcome development, as the shunt size is reduced or abolished; however, these aneurysms have been reported to cause several complications. These include tricuspid insufficiency, aortic valve prolapse, aortic insufficiency, rupture into right ventricle, right intraventricular gradient, and right ventricular outflow tract obstruction.[6-10] The size of aneurysm correlates with the severity of obstruction across outflow tract.[11] In addition, these patients remain at risk of complications of isolated VSD, such as arrhythmias, bundle branch block, aortic cusp prolapse, and infective endocarditis. During a median follow-up of 40-months, You et al. reported conduction abnormalities in 25% of patients, embolic stroke in 14%, and thrombi in 11.6% of cases with AMVS.[1] In the present case, although the aneurysm was relatively large in size, it didn't cause right intraventricular gradient or right ventricular outflow tract obstruction. In one of the largest series of AMVS, Bernardo et al. recommended closure of even a small VSD in the presence of an aneurysm along with resection or imbrication of the sac.[11] There is ~~a~~ potential of progressive enlargement of the aneurysm or other late complications. Therefore, if a conservative management strategy is considered for an asymptomatic patient, regular TTE follow-up is required for early detection of complications.

Our patient has a secundum ASD which can typically be closed by using a closure device percutaneously. However, with an associated large AMVS, the surgical closure of ASD and VSD along with aneurysmectomy and imbrication of sac were advised for him.

Conclusion

We present an unusual case of AMVS. It can be readily diagnosed by TTE. Many patients may be asymptomatic. Although both the patient and physician may be reassured by the apparent spontaneous closure of a VSD, AMVS can lead to several complications, and surgical intervention may be required despite complete closure of the VSD by the aneurysm.

Disclosure

The authors declared no conflicts of interest. Written consent was obtained from the patient/kin of the patient.

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