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RUPTURE SINUS OF VALSALVA WITH AN UNUSUAL ASSOCIATION: CASE REPORT

Dr. Vivek Gollahalli Chandrashekar MD, DM, *Dr. Natraj Setty, H.S. MD, DM, FACC, FRCP, FSCAI, Dr. Kapil Rangan MD, DM, Dr. Rama C MD, DM, Dr. Beeresh Puttaswamy MD, DM, Dr. Santhosh Jadav MD, DM, Dr. Yeriswamy M. C MD, DM, Dr. Sridhar.L. MD, DM and Dr. Manjunath, C. N. MD, DM.

Sri Jayadeva Institute of Cardiovascular Sciences and Research Bangalore

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ABSTRACT

We report a case of a 35-year-old man presenting with shortness of breath and palpitation on exertion, who was evaluated and found to have a Sub aortic membrane with severe left ventricular obstruction associated with a Right sinus of valsalva aneurysm (RSOV) rupturing into right ventricular outflow tract & perimembranous ventricular septal defect (VSD). A sinus of Valsalva aneurysm is a rare cardiac anomaly that may be congenital or acquired; a coexisting cardiac lesion might be present. Ventricular septal defect can be seen commonly associated with RSOV. However, Sub aortic membrane associated with RSOV is extremely rare. Here, we report for, an unusual presence of Sub aortic membrane with RSOV (Rupturing from right coronary sinus into right ventricular outflow tract) and Perimembranous ventricular septal defect.

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INTRODUCTION

Rupture of the sinus of Valsalva (RSOV) is an uncommonly encountered condition. Ventricular septal defect can be seen in up to 40% of cases with RSOV.[1] Its occurrence can be explained embryologically due to incomplete fusion of the aortopulmonary septum and the interventricular septum leading to RSOV and ventricular septal defect. [3, 4] Sub aortic membrane is being considered now an acquired disease. Its association with VSD is known, however its association with RSOV coexisting with VSD is extremely rare and literature regarding this is limited.

Case Presentation: A 35 year old male, with no co-morbidities presented with dyspnoea and palpitations of NYHA II class for 2 years duration with worsening of symptoms to NYHA III since 2 weeks.

Corresponding author:** ***Dr. Natraj Setty, H.S. MD,
Sri Jayadeva Institute of Cardiovascular Sciences and Research Bangalore.

On examination, he was found to have continuous murmur in the left third intercostal space with peripheral signs of aortic run off. On investigations, he was found to have cardiomegaly on chest x-ray, ECG showed left ventricular hypertrophy and 2D Echocardiography revealed the following as illustrated in the images. Parasternal long axis view shows Subaortic memebrane and windsock deformity in right coronary cusp of aorta suggestive of RSOV along with a perimembranous ventricular septal defect (Fig 1A). Colour Doppler shows sub aortic membrane with turbulence across left ventricular outflow tract & moderate aortic regurgitation, and flow across the ventricular septal defect (*Fig 1B*). Short axis view at the level of aorta with colour Doppler shows RSOV rupturing into the right ventricular outflow tract (Fig 1C). Continuous wave Doppler across left ventricular outflow tract shows severe obstruction and moderate degree of Aortic regurgitation (*Fig 1D*). Patient was found to have Rupture of sinus of valsalva (Right coronary cusp rupturing into RVOT) and perimembranous ventricular septal defect with subaortic membrance with severe obstruction and moderate Aortic

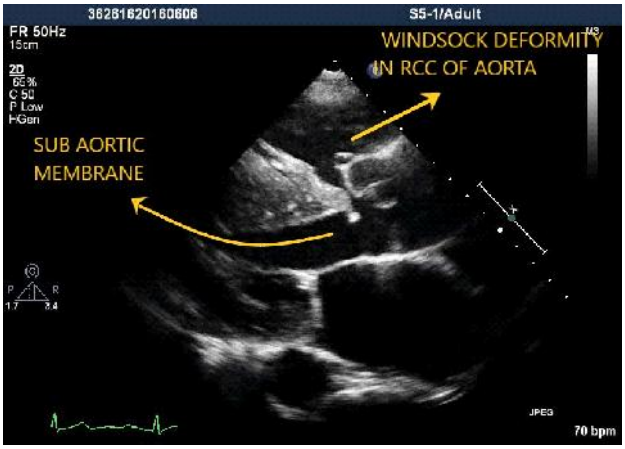

Fig 1A. Parasternal long axis view shows Subaortic memebrane and windsock deformity in right coronary cusp of aorta suggestive of RSOV along with a perimembranous ventricular septal defect

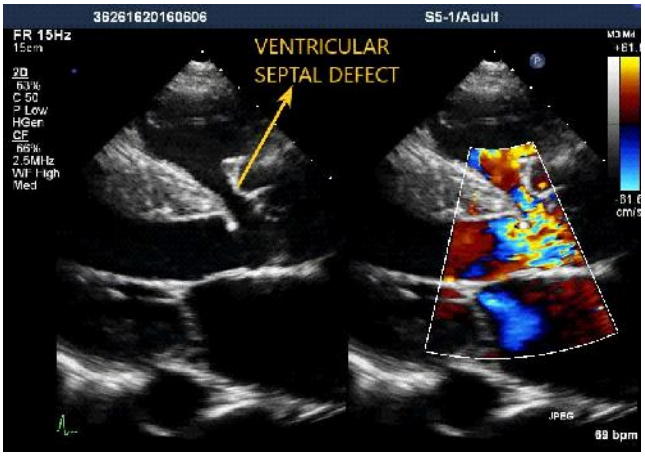

Fig 1B. Colour Doppler shows sub aortic membrane with turbulence across left ventricular outflow tract & moderate aortic regurgitation, and flow across the ventricular septal defect

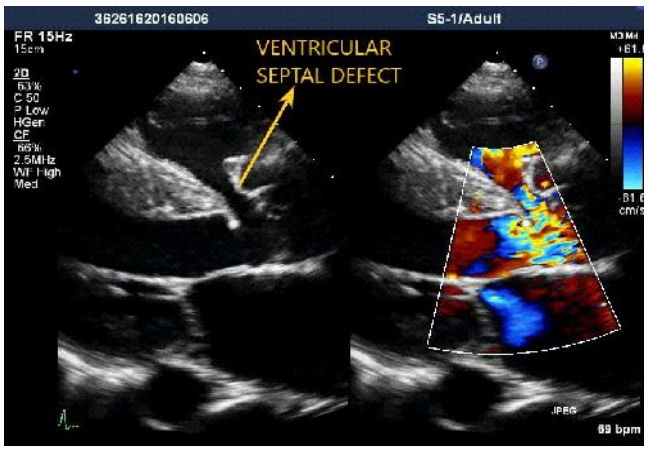

Fig 1C. Short axis view at the level of aorta with colour Doppler shows RSOV rupturing into the right ventricular outflow tract.

regurgitation. He was advised surgery, but he refused and did not give the consent for the same.

DISCUSSION

Sinus of Valsalva aneurysms is rare cardiac anomalies which may be acquired or congenital.

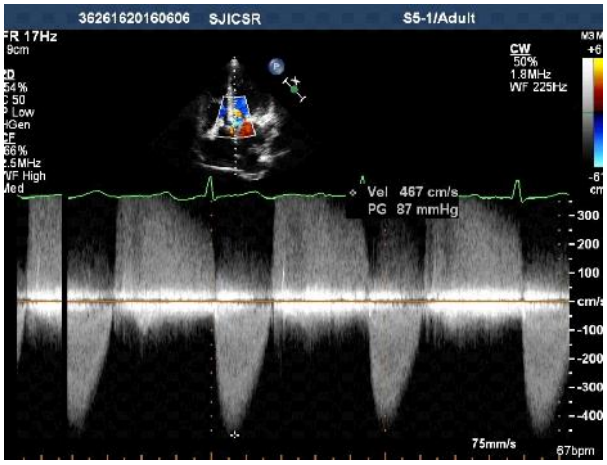

Fig 1D. Continuous wave Doppler across left ventricular outflow tract shows severe obstruction and moderate degree of Aortic regurgitation

The congenital aneurysm is more common than the acquired form.[1] Sinus of Valsalva aneurysms may occur in the right, left, or noncoronary sinus. The right is most common (75-90%), followed by the noncoronary sinus (l0-25%), with the remainder occurring in the left coronary sinus.[2] The incidence of congenital aneurysm ranges from 0.1 to 3.5% of all congenital heart defect.[3] Many investigators postulate that the defect results from the incomplete fusion of the aortopulmonary septum and the interventricular septum, the base of which forms the right and noncoronary sinuses of Valsalva. The resulting defect is a congenital weakness of the supporting tissue along the aortic annulus corresponding to the right aortic sinus and right half of the noncoronary sinus.[3,4]This is closely related to occurrence of defects in the membranous ventricular septum which is approximately 40% in congenital aneurysms.[3,5] Subvalvular aortic stenosis (SAS) is the second most common type of aortic stenosis, accounting for 14% of left ventricular outflow tract (LVOT) obstruction, with valvular aortic stenosis being the most common cause (70%).[6]

It encompasses a variety of anatomic lesions. Discrete SAS represents 75% to 85% of SAS cases, which is defined by a thin, crescent-shaped membrane just below the aortic valve. SAS is considered an acquired disease. It is rarely diagnosed during infancy, but it often manifests in the first decade of life with features of progressive LVOT obstruction, left ventricular hypertrophy (LVH), and aortic regurgitation (AR).[7] SAS is associated with defects such as VSD, AVSD, or conotruncal anomalies in 60% of cases.[8] Subaortic stenosis occurs in the natural history of ventricular septal defect usually after the first year of life, and it is progressive and requires surgery in most cases.[9] .The subaortic stenosis developed in these cases is frequently membranous and fixed, and it may occur when the diameter of the ventricular septal defect decreases, or after spontaneous closure.[10,11] The mechanism of development of discrete SAS in VSD is still unclear, and various theories are postulated. A narrow LVOT, exaggerated aortic override, increased mitral-aortic separation, and steep atrioventricular septal angle may result in a chronic flow disturbance. These factors increase the fluid shear stress on the interventricular septum and induce an abnormal endothelial and muscle proliferation resulting in the formation of a fibromuscular ridge.[12]

Conclusion

Subvalvular aortic stenosis (SAS) is the second most common type of aortic stenosis. Sub aortic membrane associated with RSOV is extremely rare. Sub aortic membrane is being considered as an acquired disease. We report a case of an unusual presence of Sub aortic membrane with RSOV (Rupturing from right coronary sinus into right ventricular outflow tract) and Perimembranous ventricular septal defect.

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