Sinus of Valsalva aneurysms complicated by infective endocarditis: one rupturing into the left ventricle and the other dissecting into the interventricular septum

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Case Report

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Abstract

Background: Sinus of Valsalva aneurysm (SVA) is an uncommon cardiac abnormality and can be a serious complication of infection or trauma or after cardiac surgery or a procedure. The clinical features and symptoms are inextricably linked to the progression and site of rupture of the aneurysm.

Case presentations: We present two rare cases of ruptured SVA, both of which were associated with infective endocarditis. In one case the SVA ruptured into the left ventricle and in the other case dissected into the interventricular septum.

Conclusions: These two cases highlight the expanding clinical view of SVA complicated by infective endocarditis using echocardiography and hemodynamic assessment to guide surgical planning.

Introduction

Sinus of Valsalva aneurysm (SVA) is a cardiac structural abnormality in which there is dilation of the aortic sinus between the aortic valve annulus and the sinotubular junction. 1 SVA can be acquired or congenital. Most cases of SVA are congenital, and SVA complicated by infective endocarditis (IE) is extremely rare. As a result of the infection, the SVA has the potential for spontaneous rupture into an adjacent cardiac structure depending on its location. This report describes two rare cases of ruptured SVA, both of which were associated with IE. In one case, the SVA ruptured into the left ventricle (LV) and in the other case dissected into the interventricular septum (IVS).

Case Presentation

Case 1

The patient was a 59-year-old man who was transferred to our hospital after presenting with progressive chest tightness, dyspnea, and shortness of breath while working. Two weeks earlier, he had been treated with an antibiotic at a local hospital for suspected IE with moderate aortic regurgitation. There was no relevant past medical history or significant family history.

On physical examination, he had a blood pressure of 125/84 mmHg, a pulse rate of 106 beats per minute, a respiratory rate of 16 per minute, and an oxygen saturation of 96% on room air. Cardiac auscultation revealed a continuous diastolic murmur best heard in the left second intercostal space and radiating to the carotid arteries. Laboratory investigations showed a normal blood cell count, normal serum electrolytes, and normal renal and liver function tests. Blood cultures were negative. An electrocardiogram showed sinus tachycardia.

Transthoracic echocardiography showed a slightly dilated LV with a diameter of 56 mm in end-diastole and a normal ejection fraction of 64%. A saccular left SVA (3.6*2.7*2.5 cm) was observed and was bulging into the left ventricular outflow tract (LVOT) (Fig. 1A). Color-flow Doppler showed abnormal flow

into the LVOT, suggesting a ruptured SVA (Fig. 1B, 1C). Transesophageal echocardiography (TEE) during systole and diastole confirmed an SVA that had ruptured from the left coronary sinus into the LVOT (Fig. 1D). A few small vegetations (measuring about 3 mm) were attached around the communication. An aortic regurgitant jet of large diameter was also noted in the LVOT (Fig. 1E). There was no associated cardiac anomaly.

In view of the threat of complications from the ruptured SVA, embolism of vegetation, and severe aortic regurgitation, surgery was performed. The surgery included resection of the aneurysm, removal of vegetation, and aortic valve replacement using a 25-mm biological aortic prosthesis (Fig. 1F). Intraoperative TEE confirmed that the bioprosthetic valve was functioning normally. At routine outpatient follow-up 4 weeks later, he reported no further symptoms and resolution of his cardiac murmur, and successful repair was confirmed on repeat transthoracic echocardiography.

Case 2

A 39-year-old man with no known health problems was admitted to our emergency department with a four-day history of acute-onset dyspnea and shortness of breath. He denied chest pain, palpitations, dizziness, and fever.

His blood pressure, heart rate, electrocardiogram, and hematology and biochemistry results were within the normal ranges.

Urgent echocardiography revealed a sac-like right SVA dissecting into the IVS (Fig. 2A). There was a 6-mm vegetation attached to the SVA (Fig. 2B). In diastole, the dissection in the IVS was filled with blood and expanded into the LVOT. Color-flow Doppler showed turbulent flow at the site of communication (Fig. 2C). TEE revealed multiple dissections of the IVS arising from a dilated right SVA (Fig. 2D) that communicated with the aortic root from the anterolateral aspect of the aortic annulus (Fig. 2E). A tri-leaflet aortic valve with severe aortic regurgitation was noted (Fig. 2F). There was no associated atrial or ventricular septal defect.

The patient subsequently underwent successful aortic valve replacement using a 23-mm mechanical valve and application of a Dacron patch. Intraoperative TEE showed that the mechanical valve was functioning normally. His postoperative course was uneventful and he was discharged in a satisfactory condition.

Discussion

SVA is a rare cardiac abnormality that is caused by weakening of the elastic lamina and muscular tissue in the aortic wall behind the sinus of Valsalva. 2,3 Most cases are congenital and secondary to connective tissue disease, including Marfan’s syndrome and many other conditions. However, acquired SVA is generally due to IE, atherosclerosis, trauma, drug or alcohol abuse, cystic medial necrosis, or degenerative disease.4 Both our patients had IE confirmed intraoperatively and by histopathology.

Infection may lead to dilatation of the sinus of Valsalva and deformation of the aortic valve cusps, culminating in rupture of the SVA and dissection of the IVS. Progression of the SVA may also increase the risk of endocarditis.5 Diagnosis of endocarditis relies mainly on the finding of vegetation and a positive blood culture. The positive blood culture rate ranges between 50% and 90% depending on the timing and quantity of blood collection, whether or not antibiotic therapy has been started. In our two cases, multiple blood cultures were negative. Positive blood cultures can help with the clinical selection of antibiotics, but negative results cannot easily rule out a diagnosis of endocarditis.

Most SVAs originate from the right coronary sinus, followed by the non-coronary sinus, and the left coronary sinus is least common.6 SVA is generally asymptomatic. However, an enlarged coronary sinus may protrude into the adjacent cardiac structure, lead to neighboring cavity obstructions, cause myocardial infarction or ischemia by compression of the coronary arteries, or, in extremely rare cases, dissect into the IVS. Strenuous activity, emotional excitement, infection, trauma, and iatrogenicity (cardiac catheterization) can induce rupture of an SVA. The majority of right SVAs rupture into the right ventricular outflow tract, followed by the right ventricle and right atrium. SVA dissecting into the IVS, as in our second case, always originates from the right coronary sinus and is usually associated with conduction abnormalities and aortic regurgitation. According to its dissecting nature, an SVA can cause congestive heart failure if it ruptures into the ventricular cavity. Less ruptured noncoronary sinus aneurysms rupture into the right atrium, right ventricle, or pericardial cavity. Left SVA typically ruptures into the LV, left atrium, or pericardial cavity, as in our first case, and is the rarest type.

The clinical features and symptoms of SVA are inextricably linked to progression of the aneurysm and the site of rupture. An unruptured aneurysm is generally clinically silent until it increases in size and compresses nearby structures or develops thromboembolic complications.7 SVA rupture is an urgent and serious situation that can manifest as sudden hemodynamic collapse, acute coronary syndrome, acute heart failure, cardiac tamponade, and even sudden death, depending on the size of the ruptured orifice, the acuity of the rupture, and the receiving chamber.8,9 As in our two cases, embolic events associated with endocarditis should be taken seriously. Coronary angiography is the gold standard for diagnosis of SVA, but was not required in our cases. Echocardiography is generally the preferred initial examination tool and can detect the location and size of the dissecting aneurysm, its relationship with adjacent structures, vegetations, and any cardiac abnormality. Vortex flow in the dissecting cavity, aortic regurgitation, and communication between the dissecting cavity and the left or right ventricle can be easily shown by color Doppler. Collapse of the aneurysm during systole, filling of the aneurysm with blood during diastole, and continuous flow in systole and diastole are useful findings in a patient with a ruptured SVA. These features were demonstrated by echocardiography in our two patients.

The differential diagnosis of SVA should include subaortic aneurysm, ventricular septal defect, coronary arteriovenous malformation, fistula, and abscess. The crucial distinction is that SVA involves structural anomalies in one or more sinuses of Valsalva in the aortic root. Echocardiographic findings are crucial for correct diagnosis of SVA, and early surgical intervention is the optimum treatment to prevent further rupture and exacerbation of symptoms. In the opinion of the authors, the prognosis of the two patients in

this report may have been better and the operation simpler if it had been performed in the absence of endocarditis.

Conclusion

In conclusion, SVA, although rare, can rupture into the LV cavity and IVS and be combined with infection, as illustrated in our two cases. Early diagnosis of SVA and surgical intervention are crucial to patient survival. Echocardiography plays a useful role in accurate diagnosis, perioperative evaluation, and follow-up.

Abbreviations

IE, infective endocarditis; IVS, interventricular septum; LV, left ventricle; LVOT, left ventricular outflow tract; SVA, Sinus of Valsalva aneurysm; TEE, transesophageal echocardiography

Declarations

Ethics approval and consent to participate

The study was approved by the Institutional Review Board at the First Affiliated Hospital of Zhejiang University (Hangzhou, China). The procedures were conducted according to the principles of the Declaration of Helsinki.

Consent for publication

Written informed consent was obtained from the patients for publication of this case report and any accompanying images. A copy of the written consent form is available for review by the Editor-in-Chief of this journal.

Availability of data and materials

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

HG wrote the manuscript. YC and YM helped to conceive the study. LX carried out the intraoperative transesophageal echocardiography. ZH and YM performed the echocardiographic examination. ZL revised the manuscript. All authors have read and approved the final manuscript.

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References

1. Bass D, Bhimji SS. Aneurysm, Sinus of Valsalva. Treasure Island (FL): StatPearls Publishing; 2018.
2. Edwards JE, Burchell HB. The pathological anatomy of deficiencies between the aortic root and the heart, including aortic sinus aneurysms. Thorax. 1957;12:125–139.
3. Ring WS. Congenital heart surgery nomenclature and database project: Aortic aneurysm, sinus of Valsalva aneurysm, and aortic dissection. Ann Thorac Surg 2000;69:S147–63.
4. Moustafa S, Mookadam F, Cooper L, et al. Sinus of Valsalva aneurysms—47 years of a single center experience and systematic overview of published reports. Am J Cardiol. 2007;99: 1159-1164.
5. Takach TJ, Reul GJ, Duncan JM, et al. Sinus of Valsalva aneurysm or fistula: management and outcome. Ann Thorac Surg 1999;68:1573-1577.
6. Bricker AO, Avutu B, Mohammed TL et al. Valsalva sinus aneurysms: Findings at CT and MR imaging. RadioGraphics. 2010 Jan;30:99-110.
7. Marques JS, Varela MG, Almeida AG, et al. Thrombosed aneurysm of the left sinus of Valsalva presenting as an intramural mass. J Am Soc Echocardiogram. 2010:23:1223.e1–1223.e3.
8. Weinreich M, Yu PJ, Trost B. Sinus of Valsalva aneurysms: review of the literature and an update on management. Clin Cardiol. 2015;38:185-189.
9. Olmedo W, Weinreich M, Villablanca PA. Ruptured sinus of Valsalva aneurysm causing myocardial infarction. Rev Esp Cardiol (Engl Ed). 2018;71:204.

Figures

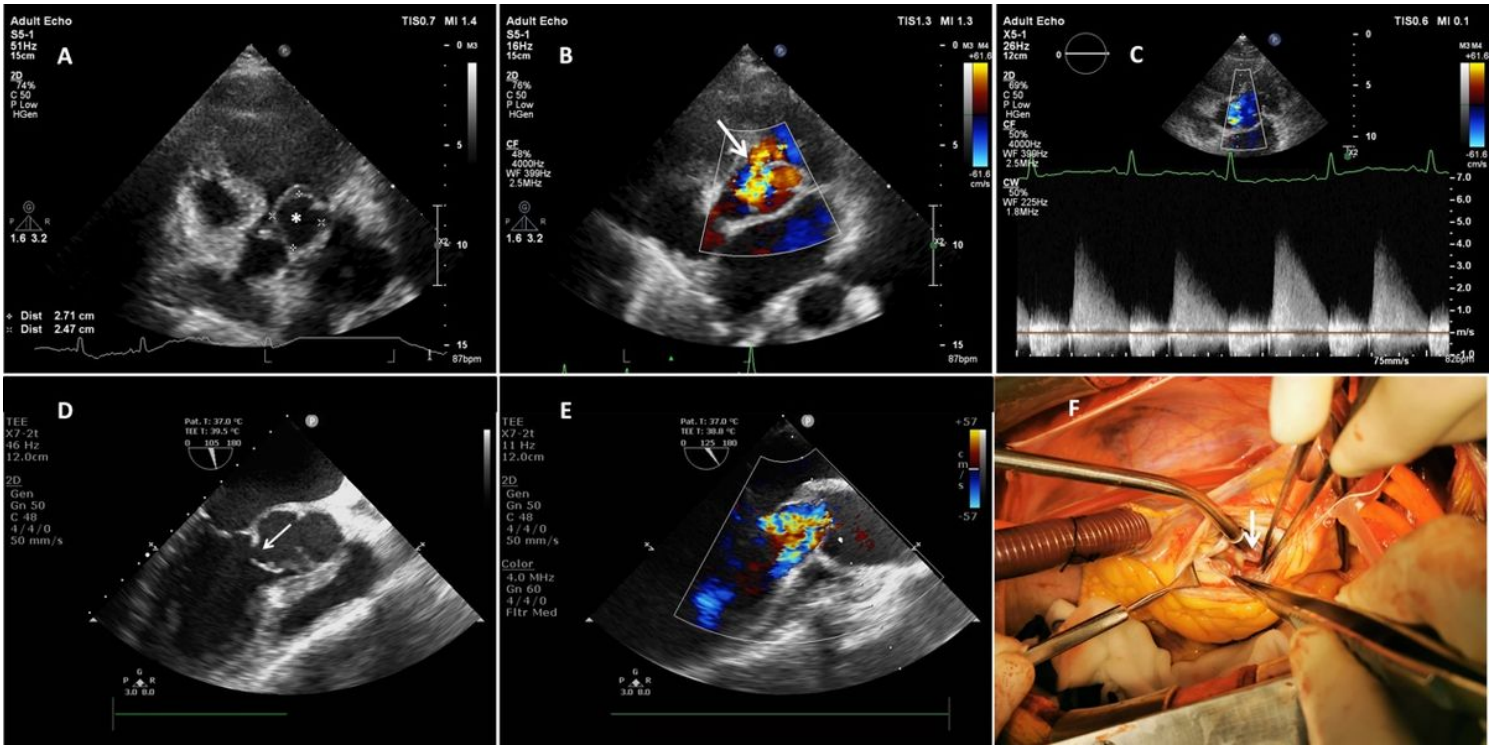

Figure 1

Rupture of a Sinus of Valsalva aneurysm in the left coronary sinus into the left ventricle. A. TTE apical five chamber view showing a left coronary SVA (*) measuring 2.7*2.5 cm and protruding into the LV. B. TTE with color-Doppler showing the flow from the left coronary SVA (arrow) into the LV. C. Pulsed-wave Doppler at the SVA. Of note is that flow is continuous but most prominently in diastole. D. Intraoperative TEE at the mid-esophageal level demonstrating the left coronary SVA with evidence of rupture (arrow). E. Intraoperative TEE at the mid-esophageal level showing a severe aortic regurgitant jet. F. Intraoperative view of the SVA inlet (arrow). LV, left ventricle; SVA, sinus of Valsalva; TEE, transesophageal echocardiography; TTE, transthoracic echocardiography

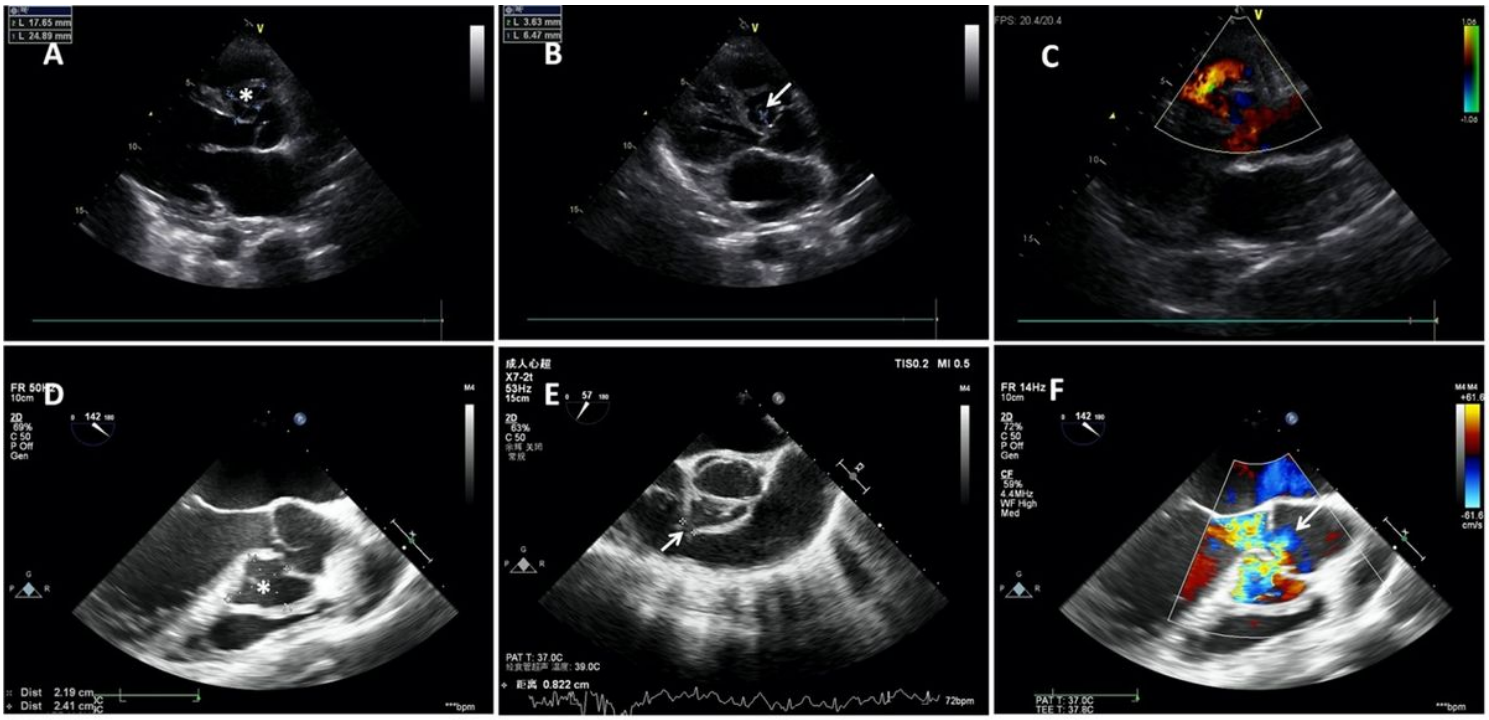

Figure 2

Rupture of a Sinus of Valsalva aneurysm in the right coronary sinus dissecting into the interventricular septum A. TTE parasternal long-axis view showing a right coronary SVA (*) measuring 1 8*2.5 cm and dissecting into the IVS. B. ATTE parasternal long-axis view noted the small vegetation (arrow) attached to the SVA.C. TTE with color Doppler showed turbulent flow at the sites of communication. D. Intraoperative TEE at the mid-esophageal level demonstrating the right coronary SVA (asterisk, *) dissecting into the IVS. E. Intraoperative TEE at the mid-esophageal level demonstrating a ruptured right coronary SVA measuring 0.8 cm at the orifice. F. Intraoperative TEE at the mid-esophageal level with color Doppler showing aortic regurgitation and the turbulent flow from the right coronary SVA (arrow) into the IVS.