

# A rare variant of a ruptured sinus of valsalva aneurysm forming an aorto-atrial fistula: a case report and review of literature

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#### **Background**

Sinus of valsalva aneurysms (SOVAs) are infrequent findings and generally diagnosed incidentally. A SOVA may be at risk for rupture, which would lead to an aorto-cardiac shunt. These patients present similarly to decompensated heart failure.

# Case Presentation

We present a case of a 44-year-old female with a ruptured non-coronary SOVA diagnosed by echocardiogram during evaluation for exertional dyspnoea. A trans-oesophageal echocardiogram (TEE) revealed a 2.1 cm non-coronary SOVA with windsock communication to the right atrium. The patient refused surgery, and two years later, presented with florid right heart failure with preserved left ventricular function. The right ventricle was severely dilated and hypokinetic with right atrial enlargement. After finally agreeing to surgery, a pre-operative catheterization revealed non-obstructive coronaries and a significant left to right shunt with elevated pulmonary pressure. The patient had suboptimal response to diuretic therapy and was sent for successful repair of the aneurysm with the closure of the aorto-atrial fistula via bovine pericardial patch and resolution of the left to right shunt as demonstrated by intra-operative TEE. Her right-sided heart failure symptoms subsequently resolved.

#### **Discussion**

SOVA is a rare finding but should still be considered in the differential in young and middle-aged patients with symptoms of acute heart failure, hemodynamic compromise, and a new continuous heart murmur. Early surgical repair is highly recommended to prevent acute and long-term complications.

#### **Keywords**

Sinus of valsalva aneurysm • Aorto-atrial fistula • Trans-oesophageal echocardiography • Case report

#### **ESC Curriculum**

2.2 Echocardiography • 6.1 Symptoms and signs of heart failure • 7.1 Haemodynamic instability • 9.1 Aortic disease

## **Learning points**

- A ruptured sinus of valsalva presents as a sudden heart failure like symptoms, such as dyspnoea on exertion, orthopnoea, and worsening lower extremity oedema.
- Echocardiography findings may show an aneurysmal outpouching of the non-coronary sinus, as well as Doppler regurgitant flow from the aortic root to the right atrium. Rupture often leads to elevated pulmonary artery pressures.
- Early surgical closure is highly recommended and can lead to rapid improvements in patient's symptoms.

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## Introduction

Sinus of valsalva aneurysm (SOVA) is a rare finding, usually diagnosed incidentally or after an aneurysm ruptures into a cardiac chamber or extracardiac structure. The estimated incidence of SOVAs in the general population is approximately 0.09% based on a large autopsy series, <sup>1</sup> of which, ruptures occur in 34%.<sup>2</sup> Out of all the ruptures, 30.0% involve the non-coronary sinus and 16% ruptured into the right atrium.<sup>3</sup> The sinus of valsalva is a region of the aortic root located between the aortic valve annulus and the sinotubular junction, and an abnormal dilatation of this region may result in a SOVA. This anomaly can be congenital (which is the most frequent aetiology) or acquired. The development of SOVAs is thought to be secondary to a weak juncture between the media and the annulus fibrosus of the aorta. <sup>4</sup> The incidence ranges from 0.1% to 3.5% of all congenital heart defects. The most common aetiologies of acquired SOVAs are trauma, bacterial endocarditis, syphilis, cystic medial necrosis, and atherosclerosis with uncontrolled hypertension.4

## **Timeline**

2 years prior	44—year-old female presented for dyspnoea on exertion. Echocardiography showed a rupture of non-coronary sinus of valsalva.
Initial presentation	Patient presented again due to severe
(Day 0)	worsening of dyspnoea, orthopnoea, and
	lower extremity oedema.
Day 6	Pre-op catheterization showing
	non-obstructive coronary arteries, elevated
	pulmonary artery pressure.
Day 8	Cardiothoracic surgery for aneurysm repair
	and aorto-atrial fistula closure.
Day 22	Patient discharged to cardiac rehab
Day 46	Repeat echocardiogram showing resolution of
	aorto-atrial fistula

# **Case presentation**

We present a case of a 44-year-old female with a ruptured non-coronary SOVA diagnosed incidentally with a trans-thoracic echocardiogram (TTE) during a routine ambulatory evaluation for dyspnoea on exertion. At that time, the TTE revealed an ejection fraction (EF) of 55–60%, normal right ventricular (RV) size and function, and a lateral tricuspid annulus peak velocity (S') of 17 cm/s. The SOVA was ill-defined on TTE. Therefore, a trans-oesophageal echocardiogram (TEE) was done, which showed a 2.3 cm non-coronary SOVA with a small windsock communication with the right atrium (*Figure 1*). The narrowest part of the aneurysmal neck appears to be 5 mm. At that time, cardiac chambers were normal in size and function with normal pulmonary pressures. The patient refused repair of the rupture and was lost to follow-up.

Two years later, she presented to our hospital with dyspnoea on exertion, orthopnoea, and progressive bilateral lower extremity oedema. On examination, she was afebrile, with a blood pressure of 150/82 mmHg and a heart rate of 89 b.p.m. Jugular vein distension was noted, and she had a continuous murmur at the third left intercostal space

and a systolic murmur over the left lower sternal border. Mild bilateral basal crackles were found on auscultation. She had three-plus bilateral pitting oedema up to the knee. A side by side comparative apical four-chamber view obtained two years apart is shown in *Figure 2*. Her ECG showed sinus rhythm, right axis deviation, and an isolated PVC. A chest X-ray showed cardiomegaly. A repeated TTE showed an EF of 50–55%, new findings of a severely dilated and hypokinetic right ventricle, right atrial enlargement, and a S' of 9 cm/s. It also showed an unchanged aneurysmal size, aorto-atrial fistula, and a continuous turbulent flow.

After agreeing to surgery, a pre-operative cardiac catheterization was obtained, which revealed non-obstructed coronaries. Right-sided catheterization measurements showed a significant left to right shunt with a pulmonary flow/systemic flow (Qp/Qs) ratio of 2.7, with a step-up of oxygenation in the right atrium and pulmonary artery, as well as an elevated pulmonary artery systolic pressure of 57 mmHg. Her pre-operative medications include aspirin, carvedilol, lisinopril, hydrochlorothiazide, and furosemide 40 mg daily. The patient had minimal response to the furosemide and was sent for a successful repair of the aneurysm with the closure of the aorto-atrial fistula via a bovine pericardial patch. Intra-operative TEE confirmed resolution of the left to right shunt. A repeated TTE showed an EF of 45–50%, along with an improvement in the RV size and function, with a S' of 13 cm/s. It also demonstrated the resolution of the fistula between the coronary sinus and the right atrium (Figure 3). After an uncomplicated post-operative course in the surgical intensive care unit, the patient was discharged with an increased dose of furosemide from 40 to 60 mg daily, along with aspirin, carvedilol, and lisinopril. Upon the 1-month clinic follow-up, clinical signs of fluid overload have resolved, and the furosemide had been discontinued.

### **Discussion**

The sinuses of valsalva are three focal expansions forming the walls of the aortic root lying between the aortic valve annulus and the sinotubular ridge. More commonly, SOVA represents a congenital anomaly, but it can also be secondary to trauma, endocarditis, syphilis, cystic medial necrosis, and atherosclerosis. Connective tissue diseases and certain infections may weaken the elastic lamina between the aortic media and the annulus fibrosis, making one more susceptible to developing a SOVA because of the high pressures subjected to this region. Congenital SOVAs can have other associated anomalies like ventricular septal defects (30–60%), aortic insufficiency (20–30%), and bicuspid aortic valves (10%). SOVAs are located at the right coronary sinus in 70% of patients, the non-coronary sinus in 10–20% and the left coronary sinus in 5%. In congenital SOVA, pathological studies have shown association with Marfan's syndrome and Ehlers-Danlos syndrome. A summary of the reported cases of SOVA is shown in (*Table 1*).

The clinical presentation of SOVA is non-specific and depends on the location, size, and if it is ruptured. Unruptured SOVAs are usually asymptomatic; however, it can present with arrhythmias due to erosion into the interventricular septum, thromboembolism originating in the aneurysm sac, or myocardial ischemia secondary to coronary compression.<sup>29</sup> SOVA rupture is the most severe complication. Symptoms vary according to the affected chamber and its size. Rupture can present as aortic regurgitation, outflow tract obstruction, arrhythmias, myocardial ischaemia, heart failure, thrombosis, stroke, or endocarditis. They can also develop aorto-cardiac shunts presenting as fatigue, chest pain, dyspnoea, or overt heart failure. <sup>2,8</sup> High clinical suspicion is important for early diagnosis and treatment. In a study done by Jung et al., 70% of patients with SOVAs had the right coronary sinus ruptured into the right ventricle, 12.5% had the right coronary sinus rupture into the right atrium, and 3.6% had the non-coronary sinus rupture into the right ventricle (similar to our patient), 30 leading to a left to right shunt, and subsequent right-sided heart failure. In our patient, we had utilized the echocardiographic parameter of the lateral tricuspic peak systolic

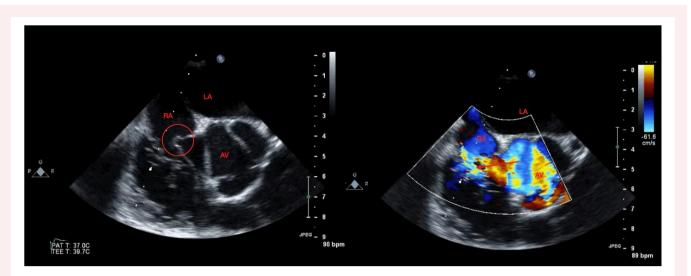


Figure 1 (A–B): A TEE mid-oesophageal short axis view at the level of the aortic valve showing the aneurysmal outpouching (windsock appearance) of the non-coronary sinus of valsalva aneurysm (circled) (A). Doppler image showing a regurgitant flow from the non-coronary sinus to the right atrium (arrow) (B). AV, Aortic Valve; LA, Left Atrium; RA, Right Atrium.



Figure 2 (A–B): Comparative TTE apical four-chamber views obtained 2 years apart, showing the natural progression of the ruptured sinus of valsalva. (A): 2 years before presentation, and (B): Obtained during presentation. LA, Left Atrium; LV, Left Ventricle; RA, Right Atrium; RV, Right Ventricle.

velocity (S') as a metric of RV systolic function. The patient had a normal S' two years prior to her acute presentation, in which the S' was found to be below normal. After the surgical intervention, the S' had improved to normal levels upon follow-up surveillance. Trans-thoracic and trans-oesophageal echocardiography are first-line imaging modalities for SOVAs. MRI and CT are useful for gaining complementary information, such as 3D anatomical reconstruction, assessment of aortic and vascular dimensions, and assessment of other case complications.<sup>31</sup>

Indications for surgery in the case of unruptured symptomatic SOVA include infective endocarditis, intractable arrhythmias, coronary artery compression, and outflow tract obstruction. Once SOVA rupture occurs, the mean survival time is 1–4 years, the mean survival time is 1–4 years, the need for early surgical repair. A retrospective study by Sarikaya et al. looked at the prognosis of 55 patients after congenital SOVA repair during a 25-year period. The prognosis at 10 and 15 years was 93% and 87%,

respectively, and the freedom from reoperation was 82% at 15 years, making surgical treatment a key in this patient's survival.<sup>4</sup>

The aetiology of this patient's ruptured SOVA is thought to be acquired, most likely secondary to long-term uncontrolled hypertension and possible atherosclerotic disease. Our patient was unique due to the non-coronary sinus being involved with rupture into the right atrium, a very atypical presentation. Fortunately, the patient had a successful surgical repair with an uneventful post-operative period, leading to a good clinical response.

SOVA is a rare finding but should still be considered in the differential in young and middle-aged patients with symptoms of acute heart failure, hemodynamic compromise, and a new continuous heart murmur. Diagnosis can be achieved non-invasively with echocardiography. Early surgical repair is highly recommended to prevent acute and long-term complications.

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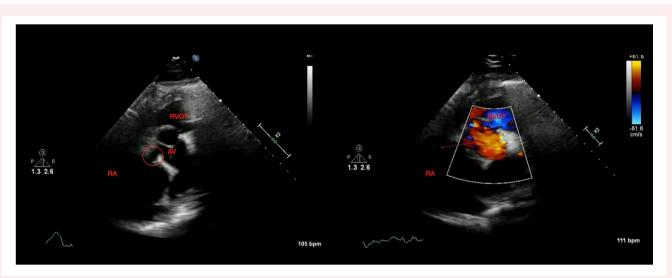


Figure 3 (A–B): A TTE parasternal short axis view at the level of the aortic valve showing the sinus of valsalva aneurysm (circled) (A). Doppler imaging (B) reveals the resolution of the regurgitant flow from the non-coronary sinus to the right atrium (arrow). AV, Aortic Valve; RA, Right Atrium; RVOT, Right Ventricular Outflow Tract.

Table 1 Cases of	sinus of valsalva aneurysms
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Author	Age (years)	Sex	SOVA characteristic	Aneurysm details	Intervention	Outcome	Notes
Ali et al. <sup>10</sup>	53	F	Ruptured non-coronary SOVA	SOVA included an aorto-atrial fistula (non-coronary cusp to right atrium)	Patch repair surgery	Recovered	
Zhuo et al. <sup>11</sup>	61	M	Ruptured right SOVA	SOVA protruded into the space between the right atrium and the visceral pericardium leading to compression of the right proximal coronary artery	Emergency excision of the right coronary sinus aneurysm, patch repair, and pericardial effusion drainage	Recovered	
Zhao et al. <sup>12</sup>	61	F	Giant ruptured non-coronary SOVA	SOVA protruded into the right atrium, completely occupying the right atrium.	Patch repair surgery	Recovered	
Deshpande et al. <sup>13</sup>	30	M	Unruptured right SOVA	Patient had two saccular aneurysms arising from the left side of the right coronary sinus without any mass effect or communication with cardiac chamber	Emergency Bentall procedure with an aortic valved conduit	Recovered	Patient was diagnosed with Takayasu arteritis.
Duval et al. <sup>14</sup>	47	F	Ruptured non-coronary SOVA	SOVA rupture into right atrium	Aneurysm was resected and sutured at its base	Recovered	An anomalous left superior vena cava was noted during surgery. The authors hypothesize that the patient suffered a

Author	Age (years)	Sex	SOVA characteristic	Aneurysm details	Intervention	Outcome	Notes
							delayed presentation of a congenital SOVA.
Doost et al. <sup>15</sup>	23	F	Ruptured right SOVA	SOVA extended into the junction between the right atrium and ventricle	Patch repair surgery	Recovered	Patient was considered to have a congenitation SOVA.
Mohammed et al. <sup>16</sup>	56	М	Unruptured left SOVA	SOVA compressed the left main stem, left anterior descending, and proximal circumflex arteries	Mechanical aortic root and ascending aortic replacement	Recovered	Authors believe the SOVA was due to degenerative disease
Ghawi et al. <sup>17</sup>	16	М	Ruptured non-coronary SOVA	Rupture into the right atrium	Patch repair surgery	Recovered	Paediatric patient had mosaic trisomy 13.
Ugurlucan et al. <sup>18</sup>	41	М	Unruptured non-coronary SOVA		Aortic root sparing surgical aortic sinus of valsalva reconstruction	Recovered	,
liang et al. <sup>19</sup>	14	F	Ruptured non-coronary SOVA	Rupture into right atrium	Percutaneous closure	Recovered	Paediatric patient presenting with vir: myocarditis with acute heart failure. Authors infer that t patient was affected by a congenital SON that ruptured due the infection.
Charfeddine et al. <sup>20</sup>	26	F	Ruptured right SOVA	Rupture into right ventricle, with a sub-aortic VSD	Patch repair surgery	The patient was monitored throughout the remainder of her pregnancy and had an uncomplicated scheduled caesarean delivery. The surgical repair was conducted after delivery and the patient fully recovered.	The patient was a pregnant woman. The authors hypothesize that the right SOVA and VS were related congenital defects due to incomplete fusion between the right and left distal bulbous septum.
Sonsoz et al. <sup>21</sup>	45	F	Unruptured non-coronary SOVA	SOVA compressed the right atrium	Patch repair surgery	Recovered	The SOVA resembled cystic cardiac mass imaging. This SOV, was deemed to be congenital.
Khanna et al. <sup>22</sup>	55	М	Unruptured non-coronary SOVA	No pressure effects	Patch repair surgery	Recovered	Thrombus formation the aneurysm mimicked a right atrial myxoma on imaging.
Mohammad et al. <sup>23</sup>	67	М	Large unruptured left SOVA		Patch repair surgery	Recovered	Patient was found to have a type 0 bicus aortic valve and type

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Author	Age (years)	Sex	SOVA characteristic	Aneurysm details	Intervention	Outcome	Notes
							R-III single coronary artery.
Ari et al. <sup>24</sup>	36	M	Ruptured non-coronary SOVA	SOVA protruded into right atrium	Repair was with a catheter-based approach with the patent ductus arteriosus occluder device	Recovered	
Hoda et al. <sup>25</sup>	16	М	Rupture of non-coronary sinus	Rupture into the right atrium	Surgical repair	Recovered	This paediatric case mimicked tricuspid valve endocarditis. No history of connective tissue disease.
Gurbuz et al. <sup>26</sup>	59	М	Unruptured left SOVA	SOVA compressed left coronary system arteries	Surgical repair	Recovered	
Prifti et al. <sup>27</sup>	52	F	Giant unruptured non-coronary SOVA	Aneurysm led to severe compression of the cardiac chambers	Surgical repair	Recovered	This was the largest unruptured aneurysm ever reported in the literature (74 × 60 mm).
Lu et al. <sup>28</sup>	52	М	Giant unruptured extracardiac right SOVA		Bentall procedure	Recovered	No connective tissue disorders; Pathological examination revealer conspicuously media mucoid degeneration of the aneurismal wa and absence of medial elastic fibres.

In accordance with COPE guidelines on publishing ethics, patient consent was not obtained due to the lack of identifying information

in the entirety of the manuscript.

# Lead author biography



John Dayco (PGY-III Internal Medicine) had overcome tremendous challenges through a passionate approach in cardiology and research. Dr Dayco is a first generation immigrant from the Philippines and also the first in his family to achieve a Bachelor's degree. Dr Dayco shares his passion for research through mentorship in the undergraduate level. As the Director of Graduate Affairs in the university's Scientific Journal Research Organization, he is actively mentoring 40-50 undergraduate

undergrad students, helping foster their youthful passion and energy

in research. Dr Dayco has accomplished this by creating a curriculum that introduces the students with the core ideas of scientific research through presentations and hosting journal clubs.

# Supplementary material

Supplementary material is available at European Heart Journal - Case Reports online.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as Supplementary data.

Consent: The authors confirm that witnessed verbal consent for submission and publication of this case report including images and associated text has been obtained from the patient in this case report. This has been discussed with the editors.

**Conflict of interest:** The authors of this article certify that there are no affiliations with any organizations or entity with any financial interest (grants, equity, ownership, etch), or non-financial interest (affiliations, personal/professional relationships) in the materials that were discussed in this manuscript.

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### Data availability

The data used upon this article are available upon request to the author for ethical and privacy reasons.

## **Ethical approval**

An institutional review board (IRB) was obtained prior to the initiation of this project. All data collections and analysis had been done to preserve patient's privacy, and no identifying information will be utilized in the entirety of this manuscript.

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