

# Surgical management for unruptured sinus of Valsalva aneurysms: a narrative review of the literature

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**Abstract:** Unruptured sinus of Valsalva aneurysms (SVAs) are rare cardiac lesions that arise due to congenital or acquired etiologies. They could be asymptomatic or cause various clinical manifestations as a consequence of their mass effect on the coronary arteries, heart valves, and other adjacent structures. While the factors predicting SVA rupture are not fully understood, ruptured SVAs carry a high complication and mortality rate, highlighting the need for early recognition and management of unruptured SVAs. Imaging modalities such as echocardiography, computed tomography (CT), angiography, and magnetic resonance imaging (MRI) are essential in identifying and characterizing the aneurysm as well as associated cardiac anomalies. However, there are no specific guidelines for the diagnosis and management of SVAs. Herein, we performed a contemporary systematic review to examine the presentation, diagnostic tests and findings, as well as outcomes for surgical intervention of unruptured SVAs. We demonstrate that surgical repair remains the preferred method of treatment in order to prevent complications such as rupture or thrombus formation. Surgery should be prompted in patients with symptomatic, large, or rapidly expanding unruptured SVAs, as well as those unruptured SVAs that contain intraluminal thrombi, have a mass effect on surrounding structures, or are recurrent. Surgical outcomes are generally good with favourable prognosis and minimal recurrence.

Keywords: Unruptured sinus of Valsalva aneurysms; sinus of Valsalva aneurysms (SVAs); surgical management

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#### Introduction

Sinus of Valsalva (SV) aneurysm (SVA) is an enlargement of the aortic root area between the aortic valve annulus and the sinotubular junction. While the factors predicting SVA rupture are not fully understood, ruptured SVAs carry a high complication and mortality rate, highlighting the need for early recognition and management of unruptured SVAs (1).

SVAs can be congenital or acquired, with the former being more prevalent (2). The true prevalence of SVAs is unknown; the

estimated rate is approximately 0.09% of the general population and 0.1% to 3.5% of all congenital cardiac defects (2). Multiple SVAs are even rarer with very few cases reported in the literature (3-5). SVAs most often affect one of the SVs, and originate predominantly from the right SV (RSV) (2,6-9). Patients with unruptured SVAs could be asymptomatic or present with non-specific symptoms such as dyspnea, chest pain, palpitation and syncope (5,10-19). Unruptured SVAs may cause valvular regurgitation, annular dilation or deformity, as well as compression of the coronary arteries, cardiac chambers and outflow tracts (20-26). A comprehensive state-of-theart imaging review of SVAs has been reported, including various imaging modalities such as echocardiography, computed tomography (CT), angiography, and magnetic resonance imaging (MRI) that can be used for the diagnosis of SVAs (24-31). SVAs may rupture into cardiac chambers or extracardiac locations, and the type of complications depends on the locations into which the rupture occurs (31). The incidence of ruptured SVAs is unknown with only a single centre review of 53 SVA cases that reported ruptured SVAs in 64% of the cases (1). While size is one criterion which may be associated with rupture, the factors predicting SVA rupture are not fully understood. Ruptured SVAs carry a high mortality rate, with a mean survival period of 3.9 years if left untreated (1).

There are no specific guidelines for the diagnosis and management of SVAs, with medical and surgical options reported (32-38). While ruptured SOVAs require urgent surgical intervention, the management of unruptured SVAs remains controversial. Unruptured SVAs that do not require surgical intervention are managed conservatively using serial follow-up surveillance imaging (31). These imaging studies provide temporal data on the size and growth of the SVA, the relationship of SVA with the surrounding structures, and potential complications. The assessment and conservative management of unruptured SVAs using multimodality cardiovascular imaging have been thoroughly discussed in a recent review and will not be reviewed in this paper (31). On the other hand, the decision to intervene on unruptured SVA is complex and multifactorial, depending on the aneurysmal sizes, the growth trends on surveillance imaging, as well as patient clinical characteristics (2). Our study focuses on the surgical management of unruptured SVAs, which is an area subject to greater controversy.

The present systematic review aims to describe the contemporary clinical presentation, diagnostic tests and findings, as well as outcomes for surgical intervention of unruptured SVAs in patients with no associated congenital heart defects, underlying connective tissue disorders or other concomitant cardiac conditions. We present the following article in accordance with the Narrative Review reporting checklist (available at http://dx.doi.org/10.21037/jtd-20-2682).

#### Methods

#### Literature search strategy

Studies examining surgical outcomes of unruptured SVAs in adult patients were identified through electronic searches

performed in June 2020 using Ovid Medline, Embase, Cochrane Database of Systematic Reviews, and Scopus. To achieve the maximum sensitivity of the search strategy, we combined the terms: "aneurysm", "aneurysms", "sinus of Valsalva", "unruptured", as well as "sinus of Valsalva aneurysms" as either keywords or MeSH terms. The reference lists of all retrieved articles were reviewed and assessed for further identification of potentially relevant studies using the inclusion and exclusion criteria. After removal of duplicates, 175 articles were screened in abstract and full-text, with 52 articles included in the final analysis (*Figure 1*).

#### Selection criteria

Eligibility of studies for the present systematic review was determined prior to commencement of data collection. These included all studies that were contemporary as defined as published after the year 2000, with described surgical management for the unruptured SVA in adult patients with no previous cardiac surgery or concomitant cardiac pathologies and no familial connective tissue diseases. All publications were limited to those involving human subjects and in the English language.

Articles were excluded if they were review articles, case reports/series on pediatric patients or published before the year 2000. Articles with insufficient information on management, those with conservative management, as well as those that described patients with familial lesions, systemic conditions, previous cardiac surgery or concomitant cardiac problems were also excluded. Papers without accessible full-texts online were also not included in our study.

#### Data extraction

All data was extracted from article texts, tables, and figures and discrepancies were resolved by group discussion and consensus.

#### Statistical analysis

Descriptive statistics were used. Non-parametric continuous variables are expressed as medians with the interquartile range (IQR). Categorical data were expressed as counts and percentages. Statistical analyses were performed using GraphPad Prism version 8.4.3, GraphPad Software, La Jolla California USA.



Figure 1 PRISMA schematic of the search strategy. PRISMA, Preferred reporting items for systematic reviews and meta-analyses.

#### **Results and discussion**

#### **Clinical presentation**

Unruptured SVAs can present at various ages, ranging from 21 to 84 years old (Table 1). The median age at presentation was 59 years old. Unruptured SVAs were more commonly reported in males (58%, 30/52 cases-patient's sex was unspecified in one case) than in females (42%, 22/52 cases). Patients with SVAs can be asymptomatic (9%, 5/53 cases) or present with non-specific symptoms such as dyspnea (43%, 23/53 cases), chest pain/pressure/tightness/discomfort (34%, 18/53 cases), palpitations (15%, 8/53 cases), and syncope/presyncope (9%, 5/53 cases). Other less common initial presentations reported include tonic clonic seizure (2%, 1/53 cases), chronic cough (2%, 1/53 cases), exertional intolerance (2%, 1/53 cases), fever (2%, 1/53 cases), dizziness (2%, 1/53 cases), orthopnea (2%, 1/53 cases), and peripheral edema (2%, 1/53 cases). More severe cases of unruptured SVAs can present with myocardial ischemia and infarction (9%, 5/53 cases), heart failure (9%, 5/53 cases), as well as cardiogenic shock (4%, 2/53 cases). Among the

patients who presented with signs and symptoms of heart failure, 4% were classified as New York Heart Association (NYHA) class II (2/53 cases), and 6% were classified as NYHA class III/IV (3/53 cases).

Murmur is a common finding (43%, 23/53 cases) on physical examination of patients with unruptured SVAs. Among these described murmurs, a diastolic murmur was appreciated in 43% of the cases (10/23 cases). A systolic murmur was also appreciated in 43% of the cases (10/23 cases), with the majority of them being described as systolic ejection murmur (30%, 7/23 cases). The majority of the reported murmurs were heard over the left sternal region (48%, 11/23 cases). Other areas such as aortic (13%, 3/23 cases), pulmonic (9%, 2/23 cases), base (4%, 1/23 cases), or mitral (4%, 1/23 cases) have also been described. Most murmurs were grade 2 or 3 (26%, 6/23 cases). Grades 1, 4, and 5 murmurs were each reported once among patients (4%, 1/23 cases). Information on the characteristics of these murmurs was scarce: 9% (2/23 cases) were described as blowing, 4% (1/23 cases) was described as soft and 4% (1/23 cases) was described as regurgitant murmur.

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## Table 1 Demographic and clinical presentation of previously described cases of sinus of Valsalva aneurysms

| Patient No | Reference                   | Voar   | Age | Sov |              |         |            | Clinica     | l presentatio | n          |                   |    | - Others             | NVHA Class  | Murmur (timing/grade/guality/location)     | Comorbidities                |
|------------|-----------------------------|--------|-----|-----|--------------|---------|------------|-------------|---------------|------------|-------------------|----|----------------------|-------------|--|------------------------------|
| Fallent NO | nelelelice                  | Tear   | Age | Sex | Asymptomatic | Dyspnea | Chest pain | Palpitation | Syncope       | Arrhythmia | Conduction defect | MI | Others               | NTTIA Class | Mumur (uming/grade/quaity/location)        | Comorbidities                |
| 1          | Pólos <i>et al</i> . (8)    | 2020   | 68  | М   | -            | Y       | -          | -           | -             | -          | -                 | -  | -                    | III–IV      | -  | Smoker                       |
| 2          | Serban et al. (9)           | 2019   | 49  | М   | -            | -       | Y          | -           | -             | -          | -                 | -  | -                    | -           | -  | _                            |
| 3          | Wang et al. (10)            | 2019   | 46  | М   | -            | Y       | -          | -           | -             | -          | -                 | -  | -                    | -           | Systolic/-/-/mitral; Diastolic/-/-/ aortic | _                            |
| 4          | Umeda <i>et al</i> . (11)   | 2018   | 69  | F   | -            | Y       | -          | -           | -             | -          | -                 | -  | -                    | -           | -  | -                            |
| 5          | Khanna <i>et al</i> . (13)  | 2017   | 55  | М   | -            | -       | Y          | -           | -             | -          | -                 | -  | -                    | -           | -  | -                            |
| 6          | Ponti <i>et al</i> . (14)   | 2017   | 71  | М   | -            | Y       | Y          | -           | -             | -          | -                 | -  | Myocardial ischemia  | -           | -  | _                            |
| 7          | Luo et al. 4                | 2017   | 48  | М   | -            | Y       | -          | -           | -             | -          | -                 | -  | -                    | -           | -  | -                            |
| 8          | Guner <i>et al</i> . (16)   | 2017   | 45  | М   | -            | Y       | Y          | -           | -             | -          | -                 | -  | -                    | -           | Early diastolic/2/blowing/L sternal border | -                            |
| 9          | Chigurupati et al. (5)      | 2017   | 39  | F   | -            | Y       | Y          | Y           | Y             | -          | AV block          | -  | -                    | -           | Diastolic/4/-/aortic                       | HTN                          |
| 10         | Giambruno et al. (17)       | 2016   | 61  | F   | Y            | -       | -          | -           | -             | -          | _                 | -  | -                    | -           | Y  | _                            |
| 11         | Prifti et al. (18)          | 2016   | 52  | F   | -            | Y       | -          | -           | -             | ST         | _                 | -  | -                    | 111         | _  | _                            |
| 12         | Sato et al. (19)            | 2016   | 75  | М   | -            | -       | -          | Y           | -             | AF         | _                 | -  | -                    | -           | _  | HTN                          |
| 13         | Qian <i>et al</i> . (12)    | 2016   | 60  | F   | -            | Y       | -          | -           | -             | -          | _                 | -  | Chest tightness      | -           | Early diastolic/-/soft/-                   | _                            |
| 14         | Karvounaris et al. (20)     | 2015   | 63  | F   | -            | Y       | -          | -           | -             | ST         | LBBB              | -  | Cardiogenic shock    | -           | Systolic/3/-/-                             | _                            |
| 15         | Gong et al. (21)            | 2015   | 45  | М   | -            | Y       | -          | -           | -             | -          | _                 | -  | -                    | -           | Diastolic/-/-/L 2nd-3rd intercostal space  | _                            |
| 16         | Chikkabasavaiah et al. (22) | 2014   | 21  | М   | -            | -       | -          | -           | -             | -          | _                 | -  | Tonic clonic seizure | -           | Early diastolic/-/-/L upper parasternal    | _                            |
| 17         | Ogiwara et al. (23)         | 2013   | 61  | F   | -            | -       | -          | -           | -             | -          | _                 | Y  | -                    | -           | _  | _                            |
|            |                             | 9y f/u | 70  | -   | -            | Y       | -          | -           | -             | -          | _                 | -  | -                    | -           | _  | _                            |
| 18         | Schönrath et al. (24)       | 2013   | 67  | М   | -            | Y       | Y          | -           | -             | -          | _                 | -  | -                    | -           | _  | _                            |
| 19         | Minagawa et al. (25)        | 2013   | 70  | М   | -            | -       | -          | -           | -             | -          | RBBB              | -  | -                    | -           | Systolic ejection/-/-/L upper parasternal  | HTN                          |
| 20         | Lu <i>et al</i> . (26)      | 2013   | 52  | М   | -            | -       | -          | -           | -             | AF         | _                 | -  | Chest discomfort     | -           | Diastolic/3/regurgitant/L sternal border   | HTN, smoker                  |
| 21         | Hu <i>et al</i> . (27)      | 2013   | 66  | F   | -            | -       | -          | Y           | -             | -          | _                 | -  | -                    | -           | _  | _                            |
| 22         | Jouni <i>et al</i> . (28)   | 2012   | 78  | Μ   | -            | Y       | Y          | -           | -             | -          | -                 | -  | -                    | -           | Diastolic/-/-/-/                           | HTN,<br>dyslipidemia,<br>OSA |
| 23         | Yagoub et al. (29)          | 2012   | 56  | М   | Y            | -       | -          | -           | -             | -          | -                 | -  | -                    | -           | Υ  | _                            |
| 24         | Saritas <i>et al</i> . (30) | 2012   | 75  | М   | _            | -       | -          | -           | Y             | -          | _                 | -  | -                    | -           | -  | _                            |
| 25         | Altarabsheh et al. (32)     | 2011   | 50  | F   | _            | -       | -          | -           | -             | -          | _                 | -  | Chronic cough        | -           | -  | _                            |
| 26         | Gupta <i>et al</i> . (33)   | 2010   | 56  | F   | _            | Y       | -          | -           | -             | -          | _                 | -  | -                    | -           | -  | _                            |
| 27         | Sohal <i>et al.</i> (34)    | 2010   | 84  | М   | _            | -       | -          | -           | Y             | -          | _                 | -  | -                    | -           | -  | _                            |
| 28         | Rosu <i>et al</i> . (35)    | 2010   | 72  | М   | _            | Y       | -          | -           | -             | -          | _                 | -  | -                    | -           | -  | _                            |
| 29         | Gunay et al. (36)           | 2010   | 36  | М   | -            | -       | -          | -           | -             |            | -                 | -  | Signs of TS          |             | -  | -                            |
| 30         | Tang and Liu (37)           | 2010   | 56  | -   | -            | -       | -          |             | _             |            | -                 | -  | -                    |             | -  | -                            |
| 31         | Bhat <i>et al.</i> (38)     | 2009   | 35  | F   | -            | -       | Y          | _           | _             |            | -                 | -  | -                    | II -        | -  | -                            |
| 32         | Matteucci et al. (39)       | 2009   | 54  | F   | -            | -       | -          | -           | Y             |            | -                 | -  | -                    | II -        | -  | HTN                          |

Table 1 (continued)

Table 1 (continued)

| Dationat Nia | Deference                  | Veer | A = - | 0   |              |         |            | Clinica     | l presentatio | n            |                      |    | Otherre                     |            |  | O a usa a da i diiti a a |
|--------------|----------------------------|------|-------|-----|--------------|---------|------------|-------------|---------------|--------------|----------------------|----|-----------------------------|------------|--|--------------------------|
| Patient No   | Reference                  | Year | Age   | Sex | Asymptomatic | Dyspnea | Chest pain | Palpitation | Syncope       | Arrhythmia   | Conduction defect    | MI | - Otners                    | NYHA Class | Murmur (timing/grade/quality/location)                                   | Comorbidities            |
| 33           | Michiels et al. (40)       | 2009 | 75    | М   | -            | Y       | -          | Y           | -             | -            | -                    | -  | -                           | -          | -  | -                        |
| 34           | Ravindranath et al. (41)   | 2009 | 35    | F   | -            | Y       | Y          | Y           | -             | -            | -                    | -  | -                           | -          | -  | -                        |
| 35           | Darabian et al. (42)       | 2009 | 32    | F   | -            | Y       | -          | -           | -             | -            | -                    | -  | -                           | -          | /-/-/blowing/L upper sternal edge  | -                        |
| 36           | Sasaki <i>et al</i> . (43) | 2009 | 56    | М   | -            | -       | -          | -           | -             | -            | -                    | -  | -                           | -          | -  | -                        |
| 37           | Yang et al. (44)           | 2008 | 69    | М   | Y            | _       | -          | -           | -             | PAC, PVC     | AV block             | -  | -                           | -          | Systolic ejection/2/-/L sternal border, 2nd and<br>3rd intercostal space | HTN                      |
| 38           | Fukui <i>et al</i> . (45)  | 2008 | 38    | F   | Y            | -       | -          | -           | -             | -            | -                    | -  | -                           | -          | Diastolic/-/-/-/   | -                        |
| 39           | Klein <i>et al</i> . (46)  | 2008 | 58    | F   | -            | -       | -          | -           | -             | -            | -                    | -  | Chest pressure              | -          | -  | -                        |
| 40           | Zannis et al. (47)         | 2007 | 24    | М   | -            | -       | Y          | Y           | -             | -            | AV block, RBBB, LBBB | -  | Exertional intolerance      | -          | -  | -                        |
| 41           | Vermeulen et al. (48)      | 2006 | 81    | F   | -            | -       | Y          | -           | -             | -            | -                    | -  | Fever                       | -          | -  | HTN, DM                  |
| 42           | Yilik <i>et al</i> . (49)  | 2006 | 29    | М   | -            | Y       | -          | -           | -             | AF           | -                    | -  | -                           | -          | -  | Smoker                   |
| 43           | Joshi <i>et al</i> . (50)  | 2006 | 78    | М   | -            | -       | Y          | -           | -             | AF           | -                    | -  | Dizziness                   | -          | Systolic ejection/-/-/pulmonic   | -                        |
| 44           | Joshi <i>et al</i> . (50)  | 2006 | 65    | М   | -            | -       | -          | -           | -             | -            | -                    | -  | -                           | -          | Y  | -                        |
| 45           | Shin <i>et al</i> . (51)   | 2005 | 35    | F   | -            | -       | Y          | -           | -             | -            | -                    | Y  | Cardiogenic shock           | -          | -  | -                        |
| 46           | Mookadam et al. (52)       | 2005 | 76    | М   | -            | Y       | -          | -           | Y             | SB           | -                    | -  | Orthopnea, peripheral edema |            | Systolic ejection/1/-/base   | HTN                      |
| 47           | Akashi <i>et al</i> . (53) | 2005 | 62    | F   | Y            | -       | -          | -           | -             | -            | AV block             | -  | -                           | -          | Systolic ejection/-/-/L parasternal                                      | -                        |
| 48           | Sharda et al. (54)         | 2004 | 38    | F   | -            | -       | Y          | Y           | -             | -            | -                    | Y  | -                           | -          | Systolic ejection/3/–/L 3rd intercostal space                            | -                        |
| 49           | Mohanakrishnan et al. (55) | 2003 | 23    | М   | -            | Y       | -          | -           | -             | EB (6–7/min) | -                    | -  | -                           | III        | Systolic ejection/-/-/L parasternal                                      | -                        |
| 50           | Banerjee and Jagasia (56)  | 2002 | 75    | М   | -            | -       | -          | -           | -             | -            | -                    | -  | -                           | -          | Diastolic/2/–/R upper sternal border                                     | -                        |
| 51           | Lijoi <i>et al.</i> (57)   | 2002 | 75    | F   | -            | -       | Y          | -           | -             | -            | -                    | -  | Myocardial ischemia         | -          | -  | HTN, smoker              |
| 52           | Rhew <i>et al</i> . (58)   | 2001 | 61    | М   | -            | -       | -          | -           | -             | PAC          | AV block, RBBB       | -  | -                           | -          | Systolic/5/–/L sternal border and pulmonic                               | -                        |
| 53           | Tsukui <i>et al</i> . (59) | 2000 | 63    | F   | -            | Y       | -          | Y           | -             | AF           | -                    | -  | -                           | -          | -  | -                        |

AF, atrial fibrillation; AV, atrioventricular; DM, diabetes mellitus; EB, ectopic beats; F, female; f/u, follow up; HTN, hypertension; LBBB, left bundle branch block; M, male; MI, myocardial infarction; N, no; NYHA, New York Heart Association; OSA, obstructive sleep apnea; PAC, premature atrial contractions; PVC, premature ventricular contractions; RBBB, right bundle branch block; SB, sinus bradycardia; ST, sinus tachycardia; TS, tricuspid stenosis; y, year; Y, yes.

#### Nguyen et al. Unruptured SVAs

Arrhythmias (21%, 11/53 cases) and conduction abnormalities (13%, 7/53 cases) were commonly reported in patients with unruptured SVAs. The most common arrhythmia observed was atrial fibrillation (9%, 5/53 cases). Other arrhythmias described include sinus tachycardia (4%, 2/53 cases), sinus bradycardia (2%, 1/53 cases), premature atrial contractions (4%, 2/53 cases), premature ventricular contractions (2%, 1/53 cases), and the presence of ectopic beats (2%, 1/53 cases). In terms of conduction issues, firstdegree atrioventricular block was reported in 9% of patients (5/53 cases), followed by right bundle branch block (6%, 3/53 cases) and left bundle branch block (4%, 2/53 cases).

Comorbidities were mentioned in 23% of patients (12/53 cases). These include hypertension (19%, 10/53 cases), obstructive sleep apnea (2%, 1/53 cases), smoking (8%, 4/53 cases), dyslipidemia (2%, 1/53 cases), and type II diabetes (2%, 1/53 cases).

#### Diagnosis

#### **Imaging modalities**

Echocardiography (92%, 49/53 cases), CT (60%, 32/53 cases), and angiography (60%, 32/53 cases) were the most frequently used imaging modalities for the diagnosis of unruptured SVAs (Table 2). Transthoracic echocardiography (TTE) (83%, 44/53 cases) offered a non-invasive initial assessment of morphology, location, and origin of an SVA. Transesophageal echocardiography (42%, 22/53 cases) was performed in cases of diagnostic uncertainty, or if involvement of the surrounding structures was not well delineated on TTE. Three-dimensional echocardiography allowed for the reconstruction of SVAs and associated lesions with excellent resolution (2%, 1/53 cases). Cardiac catheterization confirmed the diagnosis, the hemodynamic significance of the lesion, and associated cardiac abnormalities. Coronary angiography (30%, 16/53 cases), CT angiography (CTA) (25%, 13/53 cases), aortic angiography (23%, 12/53 cases), ventricular angiography (6%, 3/53 cases), and aortic CTA (2%, 1/53 cases) have all been used previously as well. CT and MRI (11%, 6/53 cases) have been used as supplemental or confirmatory tests. In some cases, the diagnosis of unruptured SVA is made intra-operatively (2%, 1/53 cases).

#### Aneurysm characteristics from imaging studies

The majority of unruptured SVA cases involved one SV (83%, 44/53 cases), although there have been cases that involved two or all three SV's (17%, 9/53 cases) (*Table 2*).

SVAs originated predominantly from the RSV (64%, 34/53 cases), followed by the non-coronary SV (NSV) (36%, 19/53 cases) and the left SV (LSV) (28%, 15/53 cases). A thrombus was present in 34% of the cases (18/53 cases). Eight percent (4/53 cases) of the reported unruptured SVAs were described as being calcified.

Unruptured SVAs may cause structural and functional anomalies of surrounding cardiac structures. Associated aortic problems such as dilation of the annulus (6%, 3/53 cases) and ascending aorta (4%, 2/53 cases) have both been described in the context of unruptured SVAs. Large unruptured SVAs can have a mass effect on adjacent cardiac chambers, outflow tracts and great vessels, thereby distorting, obstructing or compressing them (58%, 31/53 cases). The right ventricle (RV) (36%, 19/53 cases), RV outflow tract (30%, 16/53 cases), and right atrium (19%, 10/53 cases) were most commonly affected, consistent with the observation that the majority of unruptured SVAs arise from the RSV. Although less common, compression of left sided structures such as the left atrium (9%, 5/53 cases), left ventricle (LV) (6%, 3/53 cases), and LV outflow tract (2%, 1/53 cases) was also reported in the presence of unruptured SVAs, with aortic root compression and pulmonary artery constriction uncommonly reported (2%, 1/53 cases).

Valvular issues were described in 60% of the cases (32/53 cases), with aortic regurgitation being the most common (49%, 26/53 cases). Mitral regurgitation (15%, 8/53 cases), tricuspid regurgitation (8%, 4/53 cases) and tricuspid annular deformity (2%, 1/53 cases) comprise the rest of the valvular complications. Unruptured SVAs can affect the coronary arteries by displacing, compressing, obstructing or stretching them (34%, 18/53 cases). The right coronary artery (19%, 10/53 cases) and left main coronary artery (15%, 8/53 cases) were predominantly affected. Complications involving other coronary arteries such as the posterior descending artery (2%, 1/53 cases), posterior left ventricular artery (2%, 1/53 cases), left anterior descending (4%, 2/53 cases), and left circumflex artery (2%, 1/53 cases) have also been described.

#### Histopathology

Histopathological information was available in 18 case reports. Of these cases, inflammatory causes, which manifested as either inflammatory cell infiltration or nonspecific chronic inflammatory changes, were reported in 33% of patients (6/18 cases). Degenerative changes in the tunica media of the aneurysm wall were found in 44% of

| Table        | 2 Diagnostic finding:                  | s in previously descr     | ribed cases o       | of sinus of Va | lsalva aneurysms.                               |             |                           |                         |  |                                 |                                   |
|--------------|--|---------------------------|---------------------|----------------|---|-------------|---------------------------|-------------------------|--|---------------------------------|-----------------------------------|
|              |  |                           |                     | Aneurysm       | ı characteristics                               |             |                           |                         | Associate                                  | ed findings                     |                                   |
| Patier<br>No | nt<br>Reference                        | Year Diagnostic methods   | Sinus of<br>origin  | Size (mm)      | Thrombus<br>[presence/size Ca<br>(mm)/location] | Icification | Aortic<br>annulus<br>(mm) | Ascending<br>aorta (mm) | Valvular<br>complications                  | Coronary artery complications   | Others                            |
| -            | Polos <i>et al</i> . (8)               | 2020 TTE, CTA             | RCSRSV              | 50×51×64       | 1   | 1           | 35                        | I                       | AR (severe,                                | I                               | RV protrusion                     |
| 2            | Serban <i>et al.</i> (9)               | 2019 TTE, TEE,<br>CT      | RCSRSV              | 53×51          | Y/-/along<br>aneurysm's<br>wall                 | I           | I                         | I                       | AR (trivial)                               | RCA obstruction                 | RV protrusion,<br>RVOT distortion |
| ю            | Wang <i>et al.</i> (10)                | 2019 TTE                  | NCSNSV              | 36×47×51       | ≻   | I           | I                         | 54                      | MR<br>(moderate),                          | I                               | I                                 |
| 4            | Umeda <i>et al.</i> (11)               | 2018 TTE, CA              | RCSRSV              | 20×13          | Y/20/near<br>RCC–LCC<br>commissure              | I           | I                         | I                       | AR (severe)<br>MR (severe,<br>P3 prolapse) | I                               | I                                 |
| 5            | Khanna <i>et al</i> . (13)             | 2017 CA, CT               | NCSNSV              | I              | I   | I           | I                         | I                       | I  | I                               | I                                 |
|              |  | 2y f/u TTE, CT            | NCSNSV              | 28×29          | Y/18×20/<br>protruding into<br>RA               | I           | I                         | I                       | AR (mild)                                  | I                               | I                                 |
| 9            | Ponti <i>et al</i> . (14)              | 2017 CA, TTE,<br>CTA      | <b>LCSLSV</b>       | I              | I   | I           | I                         | I                       | AR (mild)                                  | LM compression                  | I                                 |
| 7            | Luo et al. (15)                        | 2017 TTE, TEE,<br>3DE CT  | NCSNSV              | 98×62×76       | I   | I           | I                         | I                       | MR (mild/                                  | I                               | LA, RA                            |
| 00           | Guner <i>et al</i> . (16)              | 2017 TTE, CT              | LCSLSV,<br>RSV, NSV | 20             | I   | I           | I                         | I                       | AR (mild)                                  | I                               | compression                       |
| 6            | Chigurupati                            | 2017 CTA, TTE             | RCSRSV,             | 41×36 (R),     | I   | I           | 22                        | 23                      | AR (severe),                               | I                               | I                                 |
| 10           | et al. (ɔ)<br>Giambruno<br>et al. (17) | 2016 TTE, CT,<br>CTA      | RCSRSV              | 59×56          | I   | I           | I                         | I                       | AR (moderate)                              | RCA ran across<br>SVA's surface | I                                 |
| #            | Prifti <i>et al.</i> (18)              | 2016 TTE, TEE,<br>CT      | NCSNSV              | 74×60          | I   | I           | I                         | I                       | I  | I                               | RA compression                    |
| 12           | Sato <i>et al.</i> (19)                | 2016 TTE, CT              | RCSRSV              | 20             | I   | I           | I                         | I                       | AR (mild)                                  | I                               | I                                 |
| 13           | Qian et al. (12)                       | 2016 TEE, CTA             | <b>LCSLSV</b>       | 87             | I   | I           | I                         | I                       | I  | I                               | LV compression                    |
| 14           | Karvounaris<br><i>et al.</i> (20)      | 2015 TTE, TEE             | LCSLSV              | 59×92          | ≻   | I           | I                         | I                       | MR<br>(moderate),<br>TR (moderate)         | LM over-<br>stretched           | PA constriction, LA<br>protrusion |
| 15           | Gong <i>et al.</i> (21)                | 2015 TTE, CT,<br>CTA, TEE | LCSLSV,<br>RSV      | 57, 57         | I   | I           | I                         | I                       | AR (moderate/<br>severe)                   | I                               | I                                 |
| Table        | ; 2 (continued)                        |                           |                     |                |   |             |                           |                         |  |                                 |                                   |

| Table        | <b>2</b> (continued)                  |                                  |                      |                         |  |               |                           |                         |                           |  |  |
|--------------|---------------------------------------|----------------------------------|----------------------|-------------------------|--|---------------|---------------------------|-------------------------|---------------------------|--|--|
|              |                                       |                                  |                      | Aneurysr                | n characteristics                              |               |                           |                         | Associat                  | ed findings:                                   |  |
| Patier<br>No | 1t Reference                          | Year Diagnostic<br>methods       | c Sinus of<br>origin | Size (mm)               | Thrombus<br>(presence/size C<br>(mm)/location) | Calcification | Aortic<br>annulus<br>(mm) | Ascending<br>aorta (mm) | Valvular<br>complications | Coronary artery<br>complications               | Others   |
| 16           | Chikkabasavaiah<br><i>et al.</i> (22) | 2014 TTE, TEE,<br>CT, MRI,<br>AA | <b>LCSLSV</b>        | 100×32×60               | 1  | >             | 1                         | 1                       | AR (moderate)             | I  | Dissection into IVS<br>and LV                    |
| 17           | Ogiwara <i>et al</i> . (23)           | 2013 AA, TTE                     | LCSLSV               | I                       | I  | I             | I                         | I                       | I                         | RCA aneurysm,<br>LAD aneurysm,<br>LM stretched | I  |
|              |                                       | 9y f/u TTE, CT                   | RCSRSV,<br>NSV       | 41×25 (R),<br>55×47 (N) | ≻  | I             | I                         | I                       | AR (severe)               | I  | Aortic root<br>compression, RA,<br>LA protrusion |
| 18           | Schonrath<br>et al. (24)              | 2013 CT, TEE,<br>MRI, CA         | <b>LCSLSV</b>        | 75                      | ≻  | I             | I                         | I                       | 1                         | LM occlusion                                   |  |
| 19           | Minagawa<br><i>et al.</i> (25)        | 2013 TTE, 3D<br>CT, CT           | RCSRSV               | 33                      | I  | I             | I                         | I                       | I                         | I  | RVOT<br>compression                              |
| 20           | Lu <i>et al</i> . (26)                | 2013 TTE, TEE,<br>ACTA,<br>CTA   | RCSRSV               | 50×33                   | I  | I             | I                         | I                       | AR (moderate)             | I  | 1  |
| 21           | Hu e <i>t al.</i> (27)                | 2013 TTE, CT                     | RCSRSV               | 75×60                   | I  | I             | I                         | I                       | I                         | RCA  | RVOT   |
| 22           | Jouni <i>et al</i> . (28)             | 2012 TTE, CTA,<br>TEE            | RCSRSV               | 51                      | I  | I             | I                         | I                       | AR (moderate/<br>severe)  | compression<br>-                               | compression<br>RVOT protrusion                   |
| 23           | Yagoub <i>et al.</i> (29)             | 2012 TTE, CT,<br>CA              | RCSRSV               | 35×37×42                | I  | I             | I                         | I                       |                           | I  | RVOT<br>compression                              |
| 24           | Saritas <i>et al.</i> (30)            | 2012 TTE, CT                     | NCSNSV               | 48×40                   | Y/28/inside<br>aneurysm sac                    | I             | I                         | I                       | I                         | I  | RA compression                                   |
| 25           | Altarabsheh<br>et al. (32)            | 2011 CT, TTE,<br>CA, AA,<br>CTA  | LCSLSV,<br>RSV, NSV  | 84×70 (L),<br>35×32 (N) | ≻  | I             | I                         | I                       | AR (trivial)              | LM compression                                 | 1  |
| 26           | Gupta <i>et al.</i> (33)              | 2010 TTE, CT,<br>CTA, CA         | NCSNSV               | I                       | ≻  | ≻             | I                         | ļ                       | I                         | I  | I  |
| 27           | Sohal et <i>al.</i> (34)              | 2010 TTE, CA,<br>AA              | RCSRSV               | 62×51                   | I  | I             | I                         | I                       | TR                        | I  | RVOT obstruction                                 |
| 28           | Rosu <i>et al.</i> (35)               | 2010 CT                          | RCSRSV               | I                       | I  | I             | I                         | I                       | I                         | I  | RVOT<br>compression                              |
| 29           | Gunay <i>et al.</i> (36)              | 2010 CT, TEE                     | NCSNSV               | I                       | I  | I             | I                         | I                       | I                         | I  | RA protrusion                                    |
| 30           | Tang and Liu (37)                     | 2010 CA, CT,<br>CTA              | RCSRSV               | I                       | I  | I             | I                         | I                       | I                         | RCA<br>compression                             | I  |
| Table        | 2 (continued)                         |                                  |                      |                         |  |               |                           |                         |                           |  |  |

#### Nguyen et al. Unruptured SVAs

| Table        | 2 (continued)                               |                            |                     |                                       |  |               |                           |                         |   |                                  |   |
|--------------|---|----------------------------|---------------------|---------------------------------------|--|---------------|---------------------------|-------------------------|---|----------------------------------|---|
|              |   |                            |                     | Aneurysm                              | n characteristics                              |               |                           |                         | Associat  | ed findings                      |   |
| Patier<br>No | nt Reference                                | Year Diagnostic methods    | Sinus of<br>origin  | Size (mm)                             | Thrombus<br>(presence/size (<br>(mm)/location) | Calcification | Aortic<br>annulus<br>(mm) | Ascending<br>aorta (mm) | Valvular<br>complications                       | Coronary artery<br>complications | Others  |
| 31           | Bhat <i>et al</i> . (38)                    | 2009 TTE, LVA,<br>AA       | LCSLSV,<br>RSV, NSV | 100×60 (L),<br>30 (R), 30<br>(N)      | >  | 1             | 1                         | 1                       | 1   | LM compression                   | RVOT<br>compression                             |
| 32           | Matteucci                                   | 2009 TEE, CT               | NCSNSV              | 67                                    | I  | I             | I                         | I                       | I   | RCA displaced                    | RA compression                                  |
| 33           | et al. (39)<br>Michiels <i>et al</i> . (40) | 2009 TTE, CTA,<br>CA       | RCSRSV              | 67×48                                 | I  | I             | I                         | I                       | I   | I                                | RV protrusion                                   |
| 34           | Ravindranath                                | 2009 TTE, AA               | LCSLSV,<br>BSV NSV  | 62×35 (L)                             | ≻  | I             | I                         | I                       | MR (mild)                                       | I                                | I   |
| 35           | et al. (+ 1)<br>Darabian<br>et al. (42)     | 2009 TTE, CTA              | NCSNSV              | 75×58                                 | I  | I             | I                         | I                       | AR (moderate)                                   | I                                | LA, RA protrusion;<br>LVOT, RVOT<br>obstruction |
| 36           | Sasaki <i>et al</i> . (43)                  | 2009 Intra-                | NCSNSV              | 30×32×36                              | ~  | ≻             | I                         | I                       | TV annular                                      | I                                | RA protrusion                                   |
| 37           | Yang <i>et al</i> . (44)                    | operative<br>2008 CT, TTE  | RCSRSV              | I                                     | ≻  | I             | I                         | I                       | deformity<br>MR (mild), TR<br>(id)              | I                                | RVOT obstruction                                |
| 38           | Fukui e <i>t al</i> . (45)                  | 2008 TTE, CT               | RCSRSV              | 52                                    | I  | I             | I                         | I                       | (mila)<br>AR (severe)                           | RCA                              | I   |
| 39           | Klein <i>et al.</i> (46)                    | 2008 CT, TTE,              | RCSRSV              | 80×60                                 | I  | I             | I                         | I                       | AR (moderate)                                   | compression<br>-                 | I   |
| 40           | Zannis <i>et al</i> . (47)                  | АА<br>2007 ТТЕ, ТЕЕ,<br>СТ | LCSLSV,<br>PSV      | I                                     | I  | I             | I                         | I                       | I   | I                                | I   |
| 41           | Vermeulen                                   | 2006 TTE, CA,<br>MPI       | RCSRSV              | 50                                    | ≻  | I             | I                         | I                       | I   | RCA obstruction                  | RV protrusion                                   |
| 42           | et al. (40)<br>Yilik <i>et al</i> . (49)    | 2006 CT, TTE,<br>TTE CA    | NCSNSV              | 97×80                                 | I  | I             | I                         | I                       | Ι   | I                                | I   |
| 43           | Joshi <i>et al</i> . (50)                   | 2006 TTE, TEE,<br>MBI CA   | RCSRSV              | 42×35                                 | I  | I             | I                         | I                       | I   | RCA displaced                    | RVOT obstruction                                |
| 44           | Joshi <i>et al</i> . (50)                   | 2006 TTE, TEE,<br>CT       | RCSRSV              | 59×49                                 | I  | I             | I                         | I                       | AR (severe)                                     | RCA involved in                  | RVOT  |
| 45           | Shin <i>et al.</i> (51)                     | С1<br>2005 ТТЕ, ТЕЕ,<br>СТ | <b>LCSLSV</b>       | 30                                    | I  | I             | I                         | I                       | AR (moderate/<br>severe)                        | LM compression                   |   |
| 46           | Mookadam<br>et al. (52)                     | 2005 TTE, TEE              | RCSRSV              | 57                                    | I  | I             | Dilated                   | Dilated                 | AR (moderate)                                   | I                                | RVOT obstruction                                |
| 47           | Akashi <i>et al</i> . (53)                  | 2005 TTE, TEE,<br>AA       | LCSLSV,<br>RSV, NSV | 42×40 (L),<br>16×20 (R),<br>60×60 (N) | I  | I             | I                         | I                       | AR (trivial),<br>MR (trivial), TR<br>(moderate) | I                                | LA, RA<br>compression                           |
| Table        | 2 (continued)                               |                            |                     |                                       |  |               |                           |                         |   |                                  |   |

|                 | ,  |  |                            | Aneurysr                    | m characteristics                              |                                |                           |                               | Associa                             | ted findings  |   |
|-----------------|--|--|----------------------------|-----------------------------|--|--------------------------------|---------------------------|-------------------------------|-------------------------------------|---|---|
| Patien<br>No    | lt<br>Reference                          | Year Diagnostic<br>methods               | Sinus of<br>origin         | Size (mm)                   | Thrombus<br>(presence/size C<br>(mm)/location) | Calcification                  | Aortic<br>annulus<br>(mm) | Ascending<br>aorta (mm)       | Valvular<br>complications           | Coronary artery<br>complications  | Others                                    |
| 48              | Sharda <i>et al.</i> (54)                | 2004 TTE, CA,<br>LVA, RVA                | RCSRSV                     | 1                           | I  | 1                              | 1                         | 1                             | I                                   | PDA, PLV<br>occlusion (due to<br>thrombi)   | RVOT obstruction                          |
| 49              | Mohanakrishnan<br>et al. (55)            | 2003 TTE, MRI,<br>CT                     | RCSRSV                     | 120×30                      | ≻  | I                              | I                         | I                             | I                                   | I   | RVOT<br>compression                       |
| 50              | Banerjee and<br>Jagasia (56)             | 2002 TTE, TEE,<br>CT, CA,<br>AA          | RCSRSV                     | 20                          | ≻  | I                              | I                         | I                             | AR (mild)                           | I   | RA compression                            |
| 51              | Lijoi et al. (57)                        | 2002 LVA, AA,<br>CA, TEE                 | LCSLSV                     | 20×70                       | >  | 1                              | I                         | I                             | 1                                   | LM, LAD, LCx<br>displaced; 1st<br>Diag, 2nd Diag<br>stretched/<br>elongated<br>causing stenosis | 1   |
| 52              | Rhew <i>et al.</i> (58)                  | 2001 TTE, CT,<br>TEE, AA                 | RCSRSV                     | 100×100                     | I  | ≻                              | I                         | I                             | AR (mild)                           | I   | RVOT<br>compression                       |
| 53              | Tsukui <i>et al.</i> (59)                | 2000 CT, AA,<br>MRI                      | NCSNSV                     | 20                          | I  | I                              | Dilated                   | I                             | AR (moderate/<br>severe)            | I   | Ι   |
| 3DE, 3<br>tomog | 8D echocardiograph<br>raphy; CTA, comput | y; AA, aortic angic<br>ted tomography ar | ography; AC<br>ngiography; | CTA, aortic<br>; Diag, diag | computed tomog<br>jonal branch of L            | graphy angio<br>AD; f/u, follo | graphy; A<br>w up; IVS    | R, aortic re<br>, interventri | gurgitation; CA,<br>cular septum; L | coronary angiogra<br>, left; LA, left atriu   | aphy; CT, compute<br>n; LAD, left anteric |

| p 5 descending; LCC, left coronary cusp; LSV, left sinus of Valsalva; LCx, left circumflex; LM, left main; LV, left ventricle; LVA, left ventricle angiography; LVOT, left ventricle outflow tract; MR, mitral regurgitation; MRI, magnetic resonance imaging; N, no; NSV, non-coronary sinus of Valsalva; PA, pulmonary artery; PDA, posterior descending right ventricle angiography; RVOT, right ventricle outflow tract; STJ, sinotubular junction; SVA, sinus of Valsalva aneurysm; TEE, transesophageal echocardiography; TR, artery; PLV, posterior left ventricular; R, right; RA, right atrium; RCA, right coronary artery; RCC, right coronary cusp; RSV, right sinus of Valsalva; RV, right ventricle; RVA, tricuspid regurgitation; TTE, transthoracic echocardiography; TV, tricuspid valve; y, year; Y, yes.

cases (8/18 cases), with mucoid deposits noted in 39% of them (7/18 cases). Damage, deficiency or absence of elastic fibers was present in 28% of the reported cases (5/18 cases). Atherosclerotic degeneration was also noted in 6% of patients (1/18 cases).

#### Surgical management and outcomes

All 53 cases (100%) of unruptured SVAs were managed surgically (*Table 3*). One patient was initially managed conservatively with medical follow-up and TTE every 6 months. However, at the two-year follow-up, the unruptured SVA was shown to increase in size, with thrombus formation and mass effect on surrounding structures, necessitating surgical intervention.

#### Indications for treatment

The rationale behind surgical treatment of unruptured SVAs was mentioned in 21 cases (*Table 3*). Among these cases, the majority of unruptured SVAs were surgically managed to as a preventative measure to avoid complications such as aneurysm rupture or thrombus formation (38%, 8/21 cases with information on treatment indication). Aneurysm size, either large or rapidly increasing size, was an indication for surgical treatment in 29% of patients (6/21 cases). Other indications included symptomatic clinical presentation (14%, 3/21 cases), presence of a thrombus (14%, 3/21 cases), observation of a mass effect on adjacent structures (14%, 3/21 cases), and recurrent aneurysm after surgical resection (5%, 1/21 cases).

#### Surgical approaches

Surgical approaches for the management of unruptured SVAs were mainly dependent on aneurysm size and the presence of associated lesions (Table 3). Small aneurysms can be repaired by direct closure of the aneurysmal orifice (4%, 2/53 cases). For larger aneurysms, patch repair was preferred as direct closure may distort the anatomy of the aortic root (66%, 35/53 cases). The presence of valvular issues generally requires valve replacement/repair or annular repair. Aortic valve (AV) replacement/repair was performed in 36% of patients (19/53 cases), AV annuloplasty in 6% (3/53 cases), mitral valve (MV) replacement/repair in 8% (4/53 cases), MV annulus reconstruction in 2% (1/53 cases), and tricuspid valve repair in 2% of patients (1/53 cases). If the involvement of the unruptured SVA was extensive and the aortic root appeared distorted, full aortic root replacement (23%, 12/53 cases) or ascending aorta

replacement (4%, 2/53 cases) may be necessary. Coronary artery bypass grafting was performed in cases where one or more coronary arteries were compromised due to mass effect of the aneurysms (17%, 9/53 cases).

#### **Operative outcomes**

The majority of operations for unruptured SVAs were uneventful (96%, 51/53 cases) (*Table 3*). In-hospital mortality was reported in two patients (4% of cases), one intraoperative and the other within 48 hours post-operation due to multi-organ failure. Patients spent 4 to 21 days in hospital after surgical management of unruptured SVAs.

### Prognosis

Follow-up, ranging from five days to nine years in duration, was reported in 31 cases (*Table 3*). The majority of these cases were asymptomatic and showed obliteration of the aneurysm as well as restoration of aortic root anatomy and valvular function (94%, 29/31). One patient needed percutaneous intervention at follow-up due to a detected leak, and the aneurysm only showed partial thrombosis after surgical repair (14). At the two-month follow-up post percutaneous intervention, CTA showed almost complete thrombosis of the aneurysm lumen. One patient was reported to have recurrence of SVA at nine-years of follow-up, which required surgical intervention (23).

#### Limitations

Our study is subject to a number of limitations. Given that our review article only includes published articles, it may be subject to publication bias. In addition, in our efforts to provide a contemporary review by limiting the inclusion criteria to articles published after the year 2000, we may have excluded other less contemporary, but relevant studies. Heterogeneity in study populations is evident. We cannot account for centre-specific practices, threshold for intervention, and postoperative management that may affect the therapeutic strategies and patient outcomes of unruptured SVAs.

#### Conclusions

Unruptured SVAs are rare entities that can cause significant morbidity and devastating consequences if ruptured. Advances in cardiac imaging have made early recognition and diagnosis of unruptured SVAs possible in a less invasive manner in recent years. Regardless, the diagnosis of

Reference

Polos et al. (8)

Patient No

1

Table 3 Treatment approaches and outcomes of sinus of Valsalva aneurysms

Year

2020

Approach

Surgery

Indication for

treatment

Clinical

#### presentation aortic root replacement Elastic fibers deficiency, mucoid Serban *et al.* (9) 2019 Surgery Aneurysm size Resection and patch repair of aneurysm, CABGx1 (SVG to RCA) 2 \_ deposits 3 Wang et al. (10) 2019 Surgery Aneurysm size, Resection of aneurysm, MVR, MV annulus reconstruction, AVR, Mucoid degeneration, abscess involvement ascending aorta replacement formation, inflammatory cells of adjacent infiltration structures 4 Umeda et al. (11) 2018 Surgery Prevent systemic Patch repair of aneurysm, MV repair Fresh thrombus with fibrin, red embolization blood cells, white blood cells, platelets Khanna et al. (13) 2017 Medical f/u, -5 \_ TTE g6m

Direct closure of the opening of aneurysm, AV repair, AV annuloplasty,

Method of repair

|    |                              | 2y f/u | Surgery     | Prevent systemi<br>embolization  | ic Resection and patch repair of aneurysm   | -    | -  | - | -               | -  |
|----|------------------------------|--------|-------------|--|---|------|--|---|-----------------|--|
| 6  | Ponti <i>et al.</i> (14)     | 2017   | Surgery     | -  | Patch repair of aneurysm, CABGx3 (LIMA to LAD, SVG to LCx, SVG to ramus)                      | -    | -  | - | -               | TTE, CTA-leak at ante<br>thrombosis of aneurys   |
|    |                              | f/u    | Percutaneou | usLeak detected<br>and only partial<br>thrombosis of<br>aneurysm post<br>surgical repair | Selective catheterization through the residual neck, implantation of Amplazer septal occluder | _    | -  | - | 2 m             | CTA-almost complete  |
| 7  | Luo <i>et al.</i> (15)       | 2017   | Surgery     | -  | Bentall procedure, MVR  | -    | -  | 7 | -               | -  |
| 8  | Guner et al. (16)            | 2017   | Surgery     | -  | Cabrol procedure  | -    | -  | - | -               | -  |
| 9  | Chigurupati et al. (5)       | 2017   | Surgery     | -  | Modified Bentall procedure  | -    | -  | - | -               | -  |
| 10 | Giambruno <i>et al.</i> (17) | 2016   | Surgery     | Aneurysm size  | Resection and patch repair of aneurysm, AVR, CABGx1 (SVG to RCA)                              | -    | No specific pathologic conditions/<br>infective processes  | 5 | 1 y             | Asymptomatic; TTE-f  |
| 11 | Prifti <i>et al.</i> (18)    | 2016   | Surgery     | Prevent rupture  | Resection and patch repair of aneurysm  | -    | Mucoid deposits, loss of elastic fibers, eosinophilic infiltration   | - | 1 m, 1 y        | 1m: CTA-complete th  |
| 12 | Sato <i>et al.</i> (19)      | 2016   | Surgery     | Aneurysm size  | Patch repair of aneurysm  | -    | -  | - | 1w, 3 m, 1<br>y | 1w: CT-no leakage of<br>aneurysm size reduction<br>thrombus formation, a<br>aneurysm and the R S |
| 13 | Qian <i>et al.</i> (12)      | 2016   | Surgery     | Prevent<br>thrombus<br>formation and<br>rupture  | Resection of aneurysm, reconstruction of coronary arteries                                    | -    | Breakage of the intimal elastic<br>fiber, lymphocytic infiltration,<br>fibroplastic proliferation,<br>calcification foci and hyaline<br>degeneration with cystic<br>degeneration of the tunica media | - | -               | -  |
| 14 | Karvounaris<br>et al. (20)   | 2015   | Surgery     | Clinical   | Bentall procedure   | Dead | -  | - | -               | -  |

(20) 15 Gong et al. (21) 2015 Surgery -AV annuloplasty, aortic sinus repair, coronary artery ostia graft \_ \_

Table 3 (continued)

1844

Complications Pathology/histology

#### 1 m TTE-competent AV, no AR

Hospital Follow-up Follow-up findings

time

\_

\_

\_

\_

stay (d)

11

1 m Asymptomatic; TTE, TEE, CT-normal AV, aortic root and ascending aorta

TTE-functioning AV and MV; CTA-restoration of normal aortic root anatomy

2 y TTE, CT-enlarged SVA originated from NSV (28x29mm), with thrombus (18x20mm), protruding into RA; mild AR

> erior border of the patch used to close the aneurysm, only partial sm; readmitted for percutaneous procedure

te thrombosis of aneurysm lumen

functioning AV and good biventricular function

rombosed cavity of the previous aneurysm; 1y: TTE-mild AR

f contrast medium into the isolated aneurysm; 3m: TTE, CTon, heterogeneous echogenicity, blood flow in the aneurysm, recurrent fistula, partial recanalization between the patched SOV; 1y: TTE-significant aneurysm size reduction, no shunt flow

Table 3 (continued)

| Patient No | Reference                             | Year   | Approach | Indication for treatment | Method of repair   | Complications  | Pathology/histology   | Hospital<br>stay (d) | Follow-up<br>time | Follow-up findings   |
|------------|---------------------------------------|--------|----------|--------------------------|--|--|---|----------------------|-------------------|--|
| 16         | Chikkabasavaiah<br><i>et al.</i> (22) | 2014   | Surgery  | -                        | A sandwich device fabricated with Gortex and Teflon felt was used to close the aneurysm (Trusler's repair), gel foam was injected to facilitate clot formation in the aneurysm, AV subcommissural annuloplasty   | -  | -   | -                    | 1 m               | Asymptomatic; TTE  |
| 17         | Ogiwara <i>et al.</i> (23)            | 2013   | Surgery  | -                        | Resection and patch repair of aneurysm, LM reimplanted using button technique, CABGx2 (SVG to LAD, SVG to RCA-IMA's were too small for bypass grafting)  | -  | Mild atherosclerotic degeneration   | -                    | 9 у               | TTE, CT—recurrent<br>LA compression                          |
|            |                                       | 9y f/u | Surgery  | Recurrent<br>aneurysms   | AVR, aortic root replacement   | Unsuccessful<br>separation from<br>bypass, cardiad<br>output was not<br>maintained,<br>dead within 48h<br>post-operative |   | -                    | -                 | -  |
| 18         | Schonrath et al. (24)                 | 2013   | Surgery  | -                        | Resection of aneurysm, aortic root replacement, CABGx2 (LIMA to LAD, RIMA to LCx)  | -  | -   | -                    | -                 | -  |
| 19         | Minagawa <i>et al.</i> (25)           | 2013   | Surgery  | Prevent rupture          | Patch repair of aneurysm   | _  | -   | 21                   | 2w, 4 m           | 2w: TTE-RVOT flor<br>leakages to SVA sat<br>RVOT flow 0.6m/s |
| 20         | Lu <i>et al</i> . (26)                | 2013   | Surgery  | -                        | Bentall procedure, modified Maze III procedure (for AF)  | -  | Diffuse mucin deposits in the media of the aneurysm, absence of medial elastic fibers | -                    | 2.5 m             | Unremarkable   |
| 21         | Hu <i>et al.</i> (27)                 | 2013   | Surgery  | -                        | Aneurysm repaired with scalloped patch of wider diameter than the distance between the sinotubular ridge superiorly and the bases of aortic annulus inferiorly, creating a pseudosinus. An aortic flap was tailored around the ostium of the RCA and sewn to the patch (the flap base was the normal aortic wall, it's free edge was corresponding to the remnant edge of the patch) | -  | Mucoid degeneration in the wall of the aneurysm                                       | f —                  | -                 | CTA, TTE—function  |
| 22         | Jouni <i>et al.</i> (28)              | 2012   | Surgery  | _                        | Patch repair of aneurysm, AVR  | -  | -   | -                    | -                 | -  |
| 23         | Yagoub et al. (29)                    | 2012   | Surgery  | -                        | Valve-sparing repair of aneurysm   | -  | -   | -                    | -                 | TTE-obliteration of  |
| 24         | Saritas et al. (30)                   | 2012   | Surgery  | -                        | Patch repair of aneurysm, AVR, CABGx3  | -  | -   | -                    | _                 | -  |
| 25         | Altarabsheh<br><i>et al.</i> (32)     | 2011   | Surgery  | -                        | AVR, aortic root replacement, reimplantation of coronary buttons   | -  | -   | -                    | -                 | -  |
| 26         | Gupta <i>et al.</i> (33)              | 2010   | Surgery  | -                        | Resection of aneurysm, ascending aorta replacement, reimplantation o R coronary button   | f –  | -   | -                    | -                 | -  |
| 27         | Sohal <i>et al.</i> (34)              | 2010   | Surgery  | -                        | Resection and patch repair of aneurysm   | -  | -   | -                    | -                 | -  |
| 28         | Rosu <i>et al.</i> (35)               | 2010   | Surgery  | -                        | Patch repair of aneurysm, reimplantation of R coronary button  | -  | -   | -                    | -                 | -  |
| 29         | Gunay <i>et al.</i> (36)              | 2010   | Surgery  | -                        | Resection and patch repair of aneurysm   | -  | -   | -                    | -                 | -  |
| 30         | Tang and Liu (37)                     | 2010   | Surgery  | -                        | Patch repair of aneurysm, AVR, CABGx1 (SVG to RCA)   | -  | -   | 9                    | -                 | -  |
| 31         | Bhat et al. (38)                      | 2009   | Surgery  | -                        | Patch repair of aneurysm   | -  | Nonspecific chronic inflammation  | -                    | 9 m               | Asymptomatic; TTE  |
| 32         | Matteucci et al. (39)                 | 2009   | Surgery  | Aneurysm size            | Resection and patch repair of aneurysm   | -  | Eosinophilic infiltration of<br>aneurysmal wall                                       | -                    | 1 m               | Asymptomatic; no L   |

Table 3 (continued)

1845

TE-clot formation within aneurysm, minimal AR

ent SVA's originated from RSV and NSV; severe AR; aortic root, RA and

flow 3.1m/s; 4m: TTE, CT—further improved RVOT flow, no AR, no sac, size reduction of the SVA sac, improvement of RVOT obstruction,

ioning AV, no AR, preserved aortic geometry

n of SVA, functioning AV

ITE-near normal dimensions of 3 sinuses, normal biventricular function no LVOT obstruction

Table 3 (continued)

1846

| Patient No | Reference                            | Year | Approach | Indication for treatment                                | Method of repair  | Complications | Pathology/histology  | Hospital<br>stay (d) | Follow-up<br>time | Follow-up findings                         |
|------------|--------------------------------------|------|----------|---|---|---------------|--|----------------------|-------------------|--|
| 33         | Michiels et al. (40)                 | 2009 | Surgery  | -   | Patch repair of aneurysm, reimplantation of R coronary button   | -             | -  | -                    | -                 | -  |
| 34         | Ravindranath<br><i>et al.</i> (41)   | 2009 | Surgery  | -   | Patch repair of aneurysm  | -             | Nonspecific chronic inflammation   | -                    | -                 | TTE—near normal dir<br>wall motion abnorma |
| 35         | Darabian et al. (42)                 | 2009 | Surgery  | -   | Resection and patch repair of aneurysm, AVR, MVR  | -             | -  | -                    | 3 m               | Unremarkable                               |
| 36         | Sasaki <i>et al.</i> (43)            | 2009 | Surgery  | -   | Patch repair of aneurysm, TV repair   | -             | Aneurysmal sac filled with a<br>highly laminated and calcified<br>agglutinative thrombus, the<br>surface of the aneurysm contained<br>only a layer of elastic fibers | -                    | 1 y               | Unremarkable                               |
| 37         | Yang <i>et al.</i> (44)              | 2008 | Surgery  | Presence of<br>intraluminal<br>thrombus                 | Patch repair of aneurysm  | -             | -  | -                    | -                 | TTE—normal aortic re                       |
| 38         | Fukui <i>et al.</i> (45)             | 2008 | Surgery  | -   | Resection and patch repair of aneurysm, AVR, reimplantation of R coronary button, reconstruction of RCA | -             | Diffusely necrotized aortic media,<br>severely destroyed elastic fiber of<br>the media   | 21                   | -                 | -  |
| 39         | Klein <i>et al.</i> (46)             | 2008 | Surgery  | _   | Aortic root replacement, reimplantation of L coronary button, CABGx1 (SVG to RCA)                       | -             | -  | 4                    | -                 | -  |
| 40         | Zannis <i>et al.</i> (47)            | 2007 | Surgery  | Aneurysm<br>size, aneurysm<br>extracardiac<br>extension | Patch repair of aneurysm  | -             | -  | 8                    | 11 m              | Unremarkable                               |
| 41         | Vermeulen <i>et al.</i> (48)         | 2006 | Surgery  | -   | Patch repair of aneurysm, CABGx1 (SVG to RCA)   | -             | Thrombus material in the<br>organisation phase suggesting<br>that the origin of the aneurysm<br>was a degenerative dissection of<br>the right coronary sinus         | 7                    | -                 | TTE-unremarkable                           |
| 42         | Yilik <i>et al.</i> (49)             | 2006 | Surgery  | -   | Resection and patch repair of aneurysm  | -             | Mucoid degenaration of the tunica media, no inflammatory change  | -                    | 5d, 3 m           | TTE-normal aortic re                       |
| 43         | Joshi <i>et al.</i> (50)             | 2006 | Surgery  | Prevent rupture   | Patch repair of aneurysm  | -             | -  | 5                    | 1 y               | TTE-competent AV,                          |
| 44         | Joshi <i>et al.</i> (50)             | 2006 | Surgery  | Prevent rupture   | AVR, hemiroot replacement, reimplantation of R coronary button  | -             | Cystic medial necrosis of the<br>aortic wall with myxoid changes in<br>the valve tissue  | 7                    | 1у                | Asymptomatic; TTE-<br>aneurysm             |
| 45         | Shin <i>et al.</i> (51)              | 2005 | Surgery  | -   | Patch repair of aneurysm, AVR   | -             | -  | -                    | -                 | TTE—normal LV wall no compression          |
| 46         | Mookadam et al. (52)                 | 2005 | Surgery  | -   | Repair of aneurysm, AV repair, resection of ventricular aneurysm  | -             | -  | -                    | 6 y               | Unremarkable                               |
| 47         | Akashi <i>et al.</i> (53)            | 2005 | Surgery  | Prevent complications                                   | Valve-sparing aortic root remodeling using Yacoub procedure   | -             | -  | -                    | -                 | AA—no AR                                   |
| 48         | Sharda et al. (54)                   | 2004 | Surgery  | -   | Patch repair of aneurysm  | -             | -  | -                    | -                 | TTE-unremarkable                           |
| 49         | Mohanakrishnan<br><i>et al.</i> (55) | 2003 | Surgery  | Clinical presentation                                   | Resection and patch repair of aneurysm, RVOT reconstruction using pericardial patch                     | -             | -  | 8                    | -                 | -  |
| 50         | Banerjee and Jagasia<br>(56)         | 2002 | Surgery  | -   | Resection and patch repair of aneurysm  | -             | -  | -                    | -                 | -  |

Table 3 (continued)

imensions of all 3 sinuses, normal biventricular function, no regional alities

root, no AR

root, no AR

, no RVOT gradient

-functioning AV, no AR, root diameter 35mm, no residual aortic

I motion, functioning AV; Multislice spiral CT-good coronary flow,

## Table 3 (continued)

| Patient No | Reference                 | Year | Approach | Indication for treatment                                 | Method of repair                            | Complication | s Pathology/histology  | Hospital<br>stay (d) | Follow-up<br>time | <sup>2</sup> Follow-up findings              |
|------------|---------------------------|------|----------|--|---|--------------|--|----------------------|-------------------|--|
| 51         | Lijoi <i>et al.</i> (57)  | 2002 | Surgery  | _  | Direct closure of the opening of aneurysm   | -            | -  | -                    | 6 m               | Asymptomatic; TTE-<br>scintigraphy, exercise |
| 52         | Rhew <i>et al.</i> (58)   | 2001 | Surgery  | Prevent rupture,<br>relieve outflow<br>tract obstruction | Patch repair of aneurysm                    | -            | -  | -                    | -                 | -  |
| 53         | Tsukui <i>et al.</i> (59) | 2000 | Surgery  | Prevent rupture  | Resection and patch repair of aneurysm, AVR | -            | Mucoid degeneration of the tunica<br>media without inflammatory<br>changes | a –                  | 10 m              | Unremarkable                                 |

AA, aortic angiography; AR, aortic regurgitation; AV, aortic valve; AVR, aortic valve; AV descending; LSV, left sinus of Valsalva; LCx, left circumflex; LIMA, left internal mammary artery; LM, left ventricle; LVOT, left ve coronary artery; RSV, right sinus of Valsalva; RIMA, right internal mammary artery; RVOT, right ventricular outflow tract; SVA, sinus of Valsalva aneurysm; SVG, saphenous vein graft; TEE, transesophageal echocardiography; TTE, transthoracic echocardiography; TV, tricuspid valve; TVR, tricuspid valve; replacement; w, week; y, year.

-normal aortic root, no AR, normal LV function; Thallium se stress test-no residual ischemia

unruptured SVAs still requires a high index of suspicion, as patients can be asymptomatic or present with non-specific symptoms. Surgical repair remains the preferred method of treatment in order to prevent complications such as rupture or thrombus formation. Surgery should be prompted in patients with symptomatic, large, or rapidly expanding unruptured SVAs, as well as those unruptured SVAs that contain intraluminal thrombi, have a mass effect on surrounding structures, or are recurrent. Surgical outcomes are generally good with favourable prognosis and minimal recurrence.

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