

Case Report

How to Treat Unruptured Sinus of Valsalva Aneurysm: A Review of the Literature

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Abstract

Background: Sinus of Valsalva Aneurysm (SVA) is a rare heart defect resulting from the frailty of the elastic lamina of the aorta. SVA can be congenital or acquired and most often, it concerns the right SV.

Methods: We describe the case of a 61-year-old male patient referred to our hospital for elective aneurysm repair of the right coronary sinus of Valsalva.

Results: Endoluminal inspection highlighted an aneurysm originating in the right coronary sinus, laminating the origin of the right coronary artery. Intraoperatively, the complex asymmetric anatomy, jointly with the poor quality of the native cusps, conducted us to perform a modified Bentall procedure.

Conclusion: Isolated SVA is a rare condition. Although the optimal management for the unruptured SVA is debated, early surgical intervention is suggested due to a potential intra or extra-cardiac rupture. Different surgical approaches were described. We believe the modified Bentall procedure is safe and durable in marked eccentric aortic regurgitation cases and with a highly deformed root.

Keywords: Aortic root; Sinus of valsalva aneurysm; Modified bentall procedure; Right coronary sinus.

Introduction

The Sinus of the Valsalva Aneurysm (SVA) is an infrequent cardiac anomaly, a product of the frailty of the elastic lamina at the junction of the aortic media and the annulus fibrosus.

SVA can be congenital or acquired. When congenital, SVA is often associated with Marfan syndrome or other connective tissue diseases. Moreover, it is usually associated with other cardiac anomalies, including doubly committed juxta-arterial ventricular septal, perimembranous ventricular septal defect, coronary anomalies, bicuspid aortic valve, aortic regurgitation,

pulmonary stenosis, tetralogy of Fallot, coarctation of the aorta or atrial septal defect. Acquired forms of SVA are associated with infections (syphilis, bacterial endocarditis, and tuberculosis), systemic inflammatory diseases (Behçet disease, ankylosing spondylitis, Takayasu disease), connective tissue disorders (systemic lupus erythematosus), medial cystic necrosis, traumatic and degenerative diseases (atherosclerosis), strenuous physical activity and abuse of drugs or alcoholism [1,2]. SVAs are more frequently in men (4:1). The incidence is higher in the Asian populations, where a ruptured SVA is a simple and invariant disease entity, in contrast to the more diverse and protean disease profiles found in the Western series [3,4]. The incidence

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of unruptured SVA is around five times higher in Asians than in Westerners. The actual prevalence is unknown; the estimated rate is approximately 0.09% of the general population up to 3.5% of all congenital cardiac defects [5].

SVA most often affects one of the SVs and originates predominantly from the right SV (70% of cases) and non-coronary Sinus in 25% [6-9]. Unruptured SVAs present at a median age of 59 years.

While ruptured SVAs are an emergency, the management of unruptured SVAs remains contentious despite early surgical intervention being recommended due to life-threatening complications. At present, the patient will be operated on according to the most recent guidelines regarding ascending aortic aneurysm [10].

Case report

A 61-year-old male patient was referred to our hospital for elective aortic root surgery. He had been observed by his general practitioner for years for an aneurysm of the right coronary sinus of Valsalva and had already refused surgery. The medical history is notable for arterial hypertension, alcoholism, active smoking and ex-drug addiction. The patient experienced only intermittent palpitations and was otherwise asymptomatic. The electrocardiogram showed an ectopic atrial rhythm. During the preprocedural diagnostic work-up, Transthoracic Echocardiography (TTE) was performed. It revealed a dilated Left Ventricle (LVESD 59 mm) with a LVEF at 54%, a tricuspid aortic valve with moderate regurgitation and an aortic root measured at 62 mm. A subsequent cardiac CT scan proved the diagnosis of isolated aneurysmal dilatation of the right coronary sinus, measured up to 65 mm, with the other two sinuses also dilated at 43 and 45 mm, with a normal sino-tubular junction (Figure 1). Intraoperatively, endoluminal inspection highlights this aneurysm developed in the right coronary sinus, laminating the origin of the right coronary artery. The right coronary and the conus artery originated from the edge of the aneurysm and have been carefully separated from the dilated sinus. The complex asymmetric anatomy, together with the inadequate quality of the native

cusps, led us to perform a modified Bentall procedure with a 25 mm frameless porcine bioprosthesis (Medtronic Freestyle). Pledged 4/0 polypropylene traction sutures are placed at each aortic valve commissure, and the valve leaflets are excised, leaving a slight edge. The coronary arteries were isolated with a small button of the full-thickness aortic wall. The Freestyle bioprosthesis was fixed in the outflow chamber of the left ventricle. The left coronary artery was reimplanted, then the right one, in the respective ostia of the bioprosthesis, with a continuous 5/0 polypropylene suture. Then, the aortic graft is trimmed to a suitable distance and sutured to the native ascending aorta using a 4/0 continuous polypropylene suture. Weaning from CPB was obtained in excellent conditions. (CPB time: 74 minutes, Cross clamp time: 59 minutes). The postoperative course was characterized by a right anterior junctional ischemic stroke, with a complete resolution of neurological disorders after the introduction of Clopidogrel.

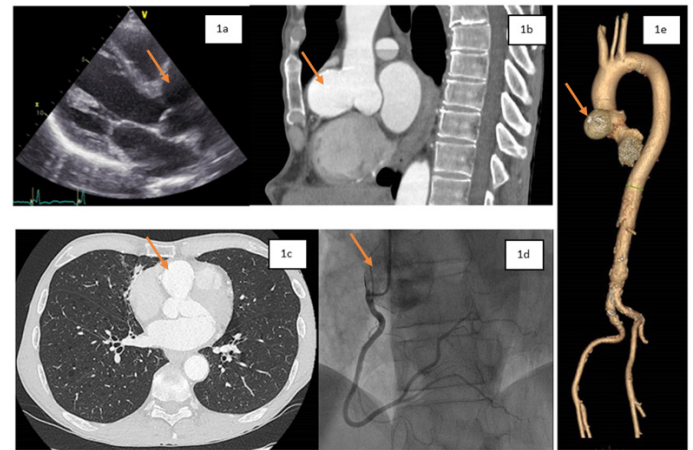


Figure 1: The aneurysmal dilatation of the right coronary sinus (arrow) on TEE at parasternal long axis view (1a); (1b). Sagittal plan of the cardiac CT (arrow on the right coronary sinus); (1c). Axial plan of the cardiac CT (arrow on the right coronary sinus); (1d). Right coronary artery with view of the right sinus of Valsalva (arrow) on coronary angiography; (1e). Three-dimensional CT displaying the aneurysm (arrow).

Table 1: Demographic, clinical presentation, treatment approaches and outcomes of sinus of valsalva aneurysm.

Reference	Year	Sex	Age	Sinus of origin	Type of Surgery	Emergency Surgery	Particularities
Fari G et al.	2020	M	58	R	None	Yes	Post mortem
Lahrouchi N et al.	2014	M	14	R	Pericardial patch	No	Down's Syndrome
Doost A et al.	2020	M	24	R	Pericardial patch	Yes	
Weinreich M et al.	2015	M	76	R	A valve-sparing aortic root replacement	No	
Weinreich M et al.	2015	F	54	NC	A valve-sparing aortic root replacement	No	
Miklos P et al.	2020	M	68	R	A remodeling technique, with a stabilization of the aortic valve annulus via running suture annuloplasty	No	
Serban AM et al.	2019	M	49	R	A tubular Dacron prosthesis; the proximal section of the tubular graft was tailored to recreate the excised right coronary sinus RCA was bypassed with a saphenous graft	No	
Abetti A et al.	2020	M	47	R	Pericardial patch	Yes	

Li X et al.	2021	M	27	L	Bentall procedure with a mechanical valve	Yes	Quadricuspid aortic valve; <i>T. pallidum</i>
Wang B et al.	2019	M	46	NC	Pericardial patch	No	
Umeda H et al.	2018	F	69	R	Gore-Tex patch	No	
Qian H et al.	2016	F	60	L	Reconstruction of the coronary arteries	No	
Ponti A et al.	2017	M	71	L	Dacron patch	No	
Luo Y et al.	2017	M	48	NC	Bentall procedure	No	
Guner A et al.	2017	M	45	R	Cabrol's technique	No	
Gong W et al.	2015	M	45	L and R	Aortic valve annuloplasty, aortic sinus repair and coronary artery ostia graft	No	
Karvounaris S et al.	2015	F	63	L	None	Yes	Obstruction of left coronary flow, ischaemic heart failure and the ongoing myocardial infarction, which led to the patient's unfavourable outcome
Ma L et al.	2022	M	51	R	Bovine epicardial patch	Yes	
P Ramirez-Rangeb et al.	2022	M	34	R	Percutaneous closure	Yes	
Petersen J et al.	2022	M	57	R	Bovine pericardial patch	No	

Discussion

The isolated Sinus of the Valsalva aneurysm is a rare condition. In a recent review of the literature, Nguyen et al. proved that patients with unruptured SVA could be asymptomatic (9%) or present with non-specific symptoms [11-17]. Arrhythmias and conduction abnormalities are common in patients with unruptured SVAs (atrial fibrillation (9%), sinus tachycardia (4%), sinus bradycardia (2%) [18]. Comorbidities include hypertension (19%), obstructive sleep apnea (2%), smoking (8%), dyslipidemia (2%) and type II diabetes (2%) [19]. The presence of the aneurysm can cause valvular regurgitation, annular dilation or deformity, and compression of the coronary arteries, cardiac chambers and outflow tracts [20-21]. Rupture often arises from a right sinus aneurysm swelling into the right ventricle, followed by a noncoronary sinus aneurysm rupturing into the right atrium [21]. In our patient, it is difficult to establish whether the aneurysm is congenital or acquired, related to the use of drugs and alcohol, and after excluding potential congenital causes. The strategy was to repair the aortic root by excision of the aneurysmal sinus with or without the aid of a pericardial patch, especially given the relatively young age of the patient and the history of drug abuse, with the risk of recurrence and, therefore, an increased risk of endocarditis with a prosthetic valve. Valve-sparing procedures (reimplantation or remodeling) could also be an option, but not in marked aortic root asymmetry cases. We could not maintain the native valve, given the marked asymmetry of the aortic root and the moderate regurgitation. In a tricuspid aortic valve with root aneurysms (root phenotype), surgery should be considered when the maximum aortic diameter is 50 mm, as recommended in recent guidelines [10]. Although optimal management for the unruptured SVA is not defined, early surgical intervention is recommended due to a potential intra or extra-cardiac rupture. Isolated SVA should be treated by surgery as soon as possible after diagnosis since they are life-threatening circumstances. Further surgical approaches were described (Table 1). The surgical strategy can be a primary closure of the aneurysm, patch repair or aortic root replacement with or without valve replacement. Direct closure may aggravate aortic regurgitation by distorting the aortic annulus and may be accountable for the recurrences. Aortic valve replacement or valvuloplasty may be necessary for aortic valve regurgitation. The long-term results of surgical repair are excel-

lent. Percutaneous treatment of an aneurysm of the Sinus of Valsalva is, in some instances, also feasible in children [22]. Aortic valve replacement is usually performed if concomitant aortic regurgitation is present [23,24].

Conclusion

Isolated SVA is a rare condition, and various surgical treatments have been described. The modified Bentall procedure is a secure and practical option in case of significant eccentric aortic regurgitation and with an extremely deformed root.

Abbreviations: CPB: CardioPulmonary bypass; CT: Computed Tomography; LVEF: left Ventricular Systolic Function; LVESD: Left Ventricular End-Systolic Diameter; SV: Sinus of Valsalva; SVA: Sinus of Valsalva Aneurysm; TTE: TransThoracic Echocardiography.

Declarations

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