Showa Univ J Med Sci 33(4), 154-158, December 2021

Case Report

A surgical case of unruptured giant sinus of Valsalva aneurysm with right ventricular outflow tract stenosis using double closure technique

Hiroshi Kataoka*[1], Hiroyuki Tanaka[1], Ayumi Omura[2], Yosuke Takei[2], Hiromoto Sone[2], Hiroshi Mase[2] and Hiroshi Suzuki[2]

Received: 26 April 2021 / Accepted: 21 July 2021

Abstract

We performed aneurysm repair with double closure technique of sinus of Valsalva aneurysm (SVA) in a 59-year-old man who was revealed obstructing right ventricular outflow tract (RVOT) with moderate Aortic insufficiency (AI) by transthoracic echocardiography (TTE). SVA plication was performed resection and suture trans tricuspid valve. Patch closure was performed with double-layer 30 mm×30 mm bovine pericardial patch. Aortic valve cusps had no abnormalities such as prolapse and AI disappeared after aneurysm repair because the etiology of AI was enlargement of aortic valve annulus. He was discharged with good condition. Computed tomography confirmed disappearance of blood flow in SVA, and TTE showed released RVOT obstruction in follow-up. Double closure technique, which combines resection and suture and patch closure, is useful for treatment of SVA.

Key words: giant sinus of Valsalva aneurysm, right ventricular outflow tract obstruction, double closure technique

Introduction

Sinus of Valsalva aneurysm (SVA) is relatively rare. No consensus has been reached regarding surgical procedure. Although various techniques including Bentall surgery and aortic repair have been reported for SVA, we have not found any report that reports the combined surgical technique of aortic aneurysm plication and patch closure. We report the case of successful surgery for a giant SVA using a double closure technique in a 59-year-old man.

Case Report

A 59-year-old man with no curreßnt complaints was referred from a local hospital for evaluation of heart murmur. He had a history of hypertension, type 2 diabetes, dyslipidemia, chronic kidney disease, hyperuricemia, and postoperative colon cancer. Transthoracic Echocardiography (TTE) revealed moderate AI with a giant SVA obstructing the RVOT. He was referred to our hospital and surgery was scheduled.

Hospital admission

Examinations of the patient upon hospital admission showed a height of 175 cm, weight 81 kg, Levine Ⅲ diastolic murmur with blood pressure of 127/73 mmHg, pulse rate of 63 per minute, and the strongest point between the fourth intercostal space and left sternal border. He was treated with Ca blocker and ARB for hypertension, statin for dyslipidemia, DPP4 inhibiter and SDLT2 inhibiter for type 2 diabetes, xanthine oxidase inhibiter for hyperuricemia.

Blood tests

Blood analysis revealed a white blood cell count of 5,630 /μl, C-reactive protein 0.08 mg/dl, total protein 7.2 g/dl, albumin 4.4 g/dl, aspartate transaminase 32 U/l, alanine transaminase 47 U/l, blood urea nitrogen 26.9 mg/dl, creatinine 1.54 mg/dl, urea acid 6.3 mg/dl,

* Corresponding author
✉ Hiroshi Kataoka
realtradedream@yahoo.co.jp

[1] Department of Cardiovascular Surgery, Showa University Fujigaoka Hospital, 1-30 Fujigaoka, Aoba-ku, Yokohama-shi, Kanagawa 227-8501, Japan.
[2] Department of Cardiology, Showa University Fujigaoka Hospital.

total cholesterol 210 mg/dl, HDL cholesterol 63 mg/dl, triglyceride 140 mg/dl, blood sugar 147 mg/dl, brain natriuretic peptide 14.1 pg/ml, and hemoglobin A1c 7.7%.

Chest X-ray

The cardiothoracic ratio was 49%. Neither pleural effusion nor lung infiltrative shadow was found. No enlargement of the cardiac shadow was found (Fig. 1A).

Electrocardiogram

The heart rate was 58 beats per minute with sinus rhythm. There was no bundle branch block or atrioventricular block. No ST-T change was showed. There was no right ventricular stress findings (Fig. 1B).

Transthoracic Echocardiography

Echocardiography revealed an SVA in the right coronary cusp (RCC) and moderate AI. Without admission of shunt flow to the right ventricle, the sinus of Valsalva protruded significantly into the right ventricle, and the RVOT was narrow (Fig. 2A, B). The aortic valve annulus diameter was 22 mm, there was no thickening or prolapse of the cusps, and the movement of the leaflets was good. No thrombus was found in the aneurysm. Tricuspid regurgitation was trivial and

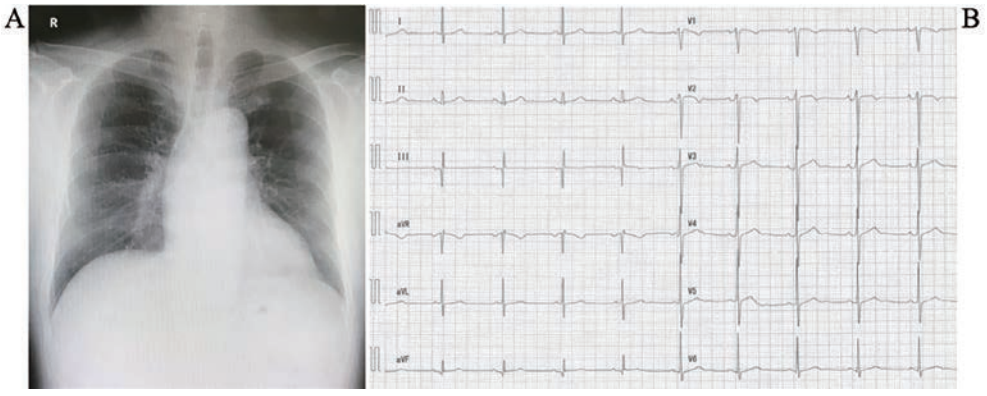

Fig. 1. Preoperative chest roentgen (A) and electrocardiogram (B)

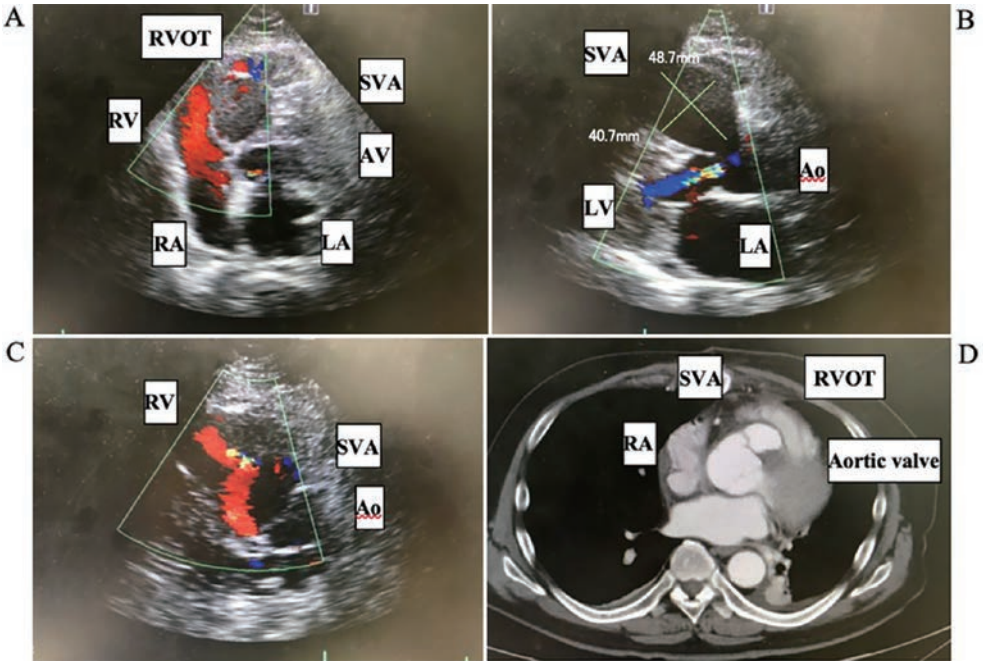

Fig. 2. Preoperative transthoracic echocardiography. The sinus of Valsalva protrudes significantly into the right ventricle, and the right ventricular outflow tract is narrow (A). The sinus of Valsalva aneurysm is 48.7 mm in diameter and has a minor axis of 40.7 mm and there is backflow into left ventricle (B). TTE about a week after operation showed blood flow into the SVA and into the right ventricle (C). CT performed at the same time showed inflow of contrast into the sutured sinus of Valsalva (D).

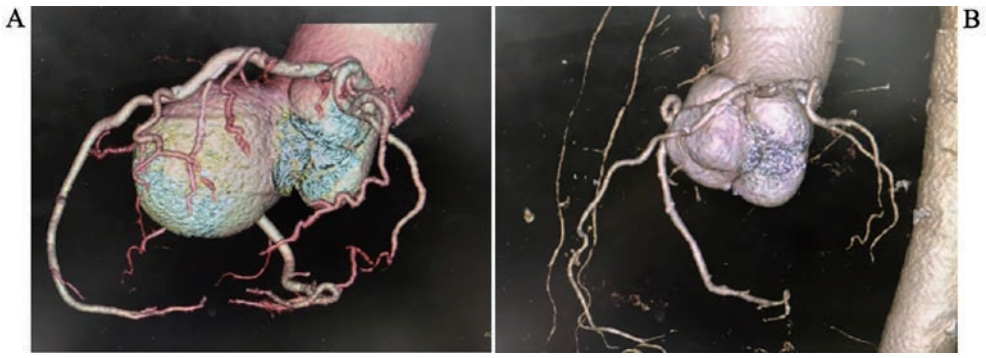

Fig. 3. 3D construction image of contrast-enhanced CT
There is SVA with aortic aneurysm of the right coronary sinus in preoperative coronary CT (A). CT showed complete thrombosis in the aneurysm and disappear of shunt flow into the right ventricle in 2-year follow up (B).

tricuspid regurgitation pressure gradient was 17 mmHg.

Computed tomography

The right coronary sinus had expanded into an aneurysm with a diameter of 71 mm and protruded into the right ventricle (Fig. 3A). There was no evidence of dilatation and dissection in the ascending aorta above the sino-tubular junction.

Operative findings

A standard median sternotomy was performed, and aortic and bicaval cannulations were used to perform cardiopulmonary bypass. The right atrium was incised after the ascending aorta was clamped, and the right ventricle was observed through the tricuspid valve. SVA of approximately 70 mm in length was protruding into the right ventricle, and ventricular septal defect or other heart disease was not confirmed. Aneurysm plication (resection and suture) was performed on the right ventricle side via tricuspid valve. The ascending aorta was transected. A 30 mm×30 mm double-layer pericardial patch (Edwards Lifesciences, Tokyo, Japan) was used to perform patch closure at the orifice of SVA on the aortic valve side. Several points were fixed with 6-0 Prolene at the lowest level of the RCC, right coronary artery entrance, and RCC and LCC borders and then a 5-0 Prolene running suture between the fixed points (Fig. 4) achieved double closure of the SVA and formation of a new oval-shaped coronary sinus. Regurgitation test confirmed the aortic valve good forms and the disappearance of AI. Operation time was 346 min and aorta closs-clamp time was 118 min, cardiopulmonary bypass time was 185 min.

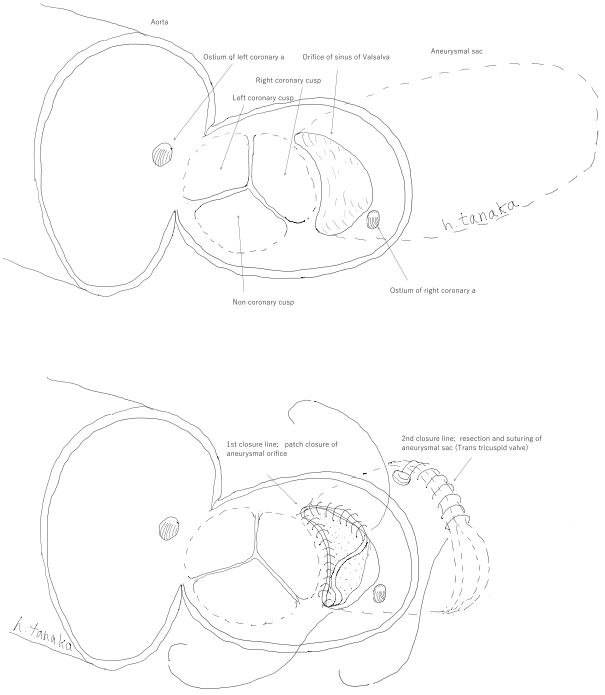

Fig. 4. Surgical procedure
First closure line perform patch closure at the orifice of SVA on the aortic valve side using a 30 mm×30 mm double-layer pericardial patch. Second closure line perform Aneurysm plication (resection and suture) trans tricuspid valve.

Postoperative course

Because the flow into the aneurysm was blocked, intraoperative transesophageal echocardiogram showed thrombus formation in the aneurysm. However, heart murmur was again present because of resumption of flow with prolongation of the coagulation system associated with warfarin medication. TTE revealed that

RVOT obstruction had disappeared, but slight shunt flow into the right ventricle (Qp/Qs 1.16) and CT showed same findings (Fig. 2C, D). Because there were no heart failure symptoms, warfarin was discontinued and we scheduled him for follow-up. He was discharged with good condition on postoperative day 10. Enhanced CT showed complete thrombosis formation in the aneurysm and disappear of shunt flow into the right ventricle in 2-year follow up (Fig. 3B). The resected SVA wall was examined pathologically. Pathological findings was left atrial tissue. There was marked nuclear deformation and enlargement of the left atrial myocardium and conspicuous fibrosis of the interstitium and fibrotic myocardial tissue. Atherosclerotic was strongly suspected, and connective tissue disease was ruled out based on physical and pathological findings.

Ethical considerations

Our institutional review board (IRB) approved this study and waived the requirement for written informed consent. The ethics approval number for this study was F2020C123at the IRB of Showa University Fujigaoka Hospital.

Discussion

SVA is a relatively rare condition that accounts for 0.15% to 1.5% of cardiopulmonary bypass cases. The gender ratio is higher for men, 3∶1 to 4∶1, with a reported average age of 45 years[1]. The causes include congenital diseases such as tetralogy of Fallot and acquired diseases related to infection, arteriosclerosis, trauma, and fragile diseases of internal tissues such as Marfan syndrome[2]. It has been reported that the incidence of coronary sinus is 70% in the right coronary sinus, 25% in the non-coronary sinus, and 5% in the left coronary sinus, with comorbid heart disease including, among others, AI, ventricular septal defect, and aortic stenosis[2, 3]. SVA often requires surgical repair because it expands into the heart and ruptures into the right atrium or right ventricle, resulting in heart failure.

However, there are recent reports suggesting that because of decreasing surgical mortality and a favorable long-term prognosis, surgery is required, even in unruptured cases, before complications such as AI, RVOT obstruction, and myocardial ischemia arise[1-4]. Wang *et al.*[4] reported that AI in particular affects the prognosis. Although we observed no sign of heart failure, we decided to perform an operation because of TTE identified moderate AI, a 70 mm aneurysm, and RVOT obstruction.

The surgical procedure is selected according to the location of the expanding SVA, the presence of rupture, and the degree of AI. SVA closure includes simple closure and patch closure. Simple closure carries the risk of relapsed aneurysm and regurgitation caused by aortic valve deformity. As reported by Wang *et al.*[4-6], residual AI has a poor prognosis often requiring aortic replacement or valvuloplasty, or aortic root replacement. Polos *et al.*[7] performed a valve-sparing root reconstruction procedure (remodeling technique) for an SVA and Carlotta *et al.*[8] conducted an aortic valve repair using a hemi-remodeling technique. However, Wingo *et al.*[9] and Abraiov *et al.*[10] reported in a retrospective study that good results can be obtained by patch closure or aneurysm plication.

Although single suture or patch closure is commonly used for SVA, we performed both plication (single suture) and patch closure of SVA to ensure the release of RVOT. Double closure of the SVA using a combination of both techniques finally resulted in the disappearance of flow. As there was no aortic valve annulus enlargement and no aortic abnormality such as thickening and cusp prolapse, we considered that AI developed because the RCC was pulled by the SVA. Thus, release of RCC traction appeared to have improved AI without aortic valve replacement (AVR). Since mechanical valve is choice for aortic valve replacement in younger patients, we were able to avoid permanent warfarin medication by not performing AVR.

Conclusion

We achieved perfect repair of SVA by double closure technique with patch closure and aneurysm plication. We were able to control AI and avoid aortic valve replacement with a mechanical valve. We conclude that double closure technique produced satisfactory results with no evidence of reexpansion of the sinus of Valsalva, appearance of shunt flow in the repaired aortic aneurysm, or AI in 2-year follow up.

Conflict of Interest Disclosure

No potential conflict of interest relevant to this article is reported.

Disclosure forms provided by the authors are available with the full text of this article at the Showa University Journal of Medical Sciences.

Disclosure Statement

We have no financial or other interest in the manufacture or distribution of the device.

Acknowledgments

We thank Nancy Schatken, BS, MT (ASCP) and Hugh McGonigle, from Edanz Group (https://en-author-services.edanz.com/ac), for editing a draft of this manuscript.

References

1. Moustafa S, Mookadam F, Cooper L, *et al*. Sinus of Valsalva aneurysm-47 years of a single center experience and systematic overview of published reports. *Am J Cardiol*. 2007;**99**:1159–1164.
2. Feldman DN, Roman MJ. Aneurysms of the sinuses of Valsalva. *Cardiology*. 2006;**106**:73–81.
3. Harkness JR, Fitton TP, Barrerio CJ, *et al*. A 32-year experience with surgical repair of sinus of Valsalva aneurysms. *J Card Surg*. 2005;**20**:198–204.
4. Wang ZJ, Zou CW, Li DC, *et al*. Surgical repair of sinus of Valsalva aneurysm in Asian patients. *Ann Thorac Surg*. 2007;**84**:156–160.
5. Takach TJ, Reul GJ, Duncan JM, *et al*. Sinus of Valsalva aneurysm or fistula: management and outcome. *Ann Thorac Surg*. 1999;**68**:1573–1577.
6. Thankachen R, Gnanamuthu R, Doshi H, *et al*. Unruptured aneurysm of the sinus of Valsalva presenting with right ventricular outflow obstruction. *Tex Heart Inst J*. 2003;**30**:152–154.
7. Polos M, Sulea CM, Benke K, *et al*. Giant unruptured sinus of Valsalva aneurysm successfully managed with valve-sparing procedure –a case report. *J Cardiothorac Surg*. 2020;**15**:6. (accessed 2020 Jan 25) Available from: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC6950913/pdf/13019_2020_Article_1061.pdf
8. Carlotta B, Zakkar M, Zacek P, *et al*. Bicuspid aortic valve repair with hemi-remodeling technique and external ring annuloplasty. *Card Surg*. 2020;**35**:146–150.
9. Wingo M, de Angelis P, Worku BM, *et al*. Sinus of Valsalva aneurysm repairs: operative technique and lessons learned. *J Card Surg*. 2019;**34**:400–403.
10. Abralov K, Alimov A. Short-term results of sinus of Valsalva aneurysm repair. *World J Pediatr Congenit Heart Surg*. 2017;**8**:13–17.