Original Article

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Repair of Ruptured Sinus of Valsalva Aneurysm: 15 years of Single Center experience

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Abstract

Background and Aims: Ruptured sinus of Valsalva aneurysm is an uncommon cardiac anomaly, fatal if not treated in time. This study was designed to retrospectively analyze our 15-year experience of the surgical repair for ruptured sinus of Valsalva aneurysm and to study the optimal surgical strategy, morbidity, mortality, and long-term surgical outcome.

Methods: This study was conducted on 48 (36 Male, and 12 Female) patients of ruptured sinus of Valsalva aneurysm, operated at department of cardiac surgery, Shahid Gangalal National Heart Centre, Nepal, from January 2006 to December 2020 and followed up till March 2021. Follow-up data were obtained from the outpatient department records and telephone calls.

Results: Mean age was 30.17±11.5 (12-63) years. Rupture of the right coronary sinus into the right atrium was the most common anatomic type (52%). Preoperative aortic regurgitation equal to or greater than grade II were seen in 9 patients (19%) and ventricular septal defects in 6 cases (13%). Two patients had preoperative renal failure. One patient had associated aortic root dilatation and underwent modified Bentall's procedure. Ruptured sinus of Valsalva aneurysm was repaired from single chamber approach in 9 patients, and dual chamber approach in 39. There was a single mortality (2.08%), two patients required permanent pacemaker placement for complete heart block, and two had wound infection. Follow-up data were available for 36 patients (75%). With the mean follow up of 7.07±3.93 (range, 0.83-15) years, there was no recurrence present. All survivors were in New York Heart Association functional Class I or II. There was one late death, due to non-cardiac cause.

Conclusion: Ruptured sinus of Valsalva aneurysm is rare, yet prompt diagnosis and optimal surgical management is crucial in reducing the deleterious effects. Surgical repair of ruptured sinus of Valsalva carries an acceptable low operative risk and can be performed with laudable long-term outcome, with low incidence of recurrence.

Keywords: Ruptured sinus of Valsalva, Ruptured aneurysm, Congenital cardiac lesions.

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Introduction

Ruptured Sinus of Valsalva aneurysm (RSOV), either congenital or acquired, is a rare cardiac anomaly. The estimated incidence of Sinus of Valsalva aneurysms (SVA) is approximately 1.24% to 4.9% in Asians, five times more frequent in Asians than in western population.[1-3] They comprise up to 3.5% of all congenital heart defects, and 0.14%-3% of all open-heart surgical procedures.[1,3]

Sinus of Valsalva aneurysms (SVA) are commonly associated with other congenital cardiac defects like ventricular septal defects (VSD), atrial septal defects (ASD) and aortic regurgitation (AR). These commonly rupture into right ventricle and right atrium but rarely into left heart chamber because of thick wall of left ventricle.[1,4]

Delay in the management of RSOVs ultimately results in heart failure and affects patient's life expectancy. The mean survival period for untreated patients is 1–2 years following rupture, which warrants the need for early surgical intervention.[4] Surgical repair has been the traditional treatment for these aneurysms. Few large or long-term series exist regarding the RSOV.

Surgical repair varies from simple plication, to single and double patch repair. In the beginning of experiences, RSOV repair was approached through the chamber into which the aneurysm ruptured, and direct suture closure of the fistula was performed. However, with this approach few patients required reoperation for recurrent rupture or residual VSD with or without aortic valve repair. Therefore, a double approach has been used in most recent patients.[5] We sought to review our 15-year surgical experience following repair of RSOV in our center and retrospectively analyze the optimal surgical strategy, operative risk and determinants of long-term surgical determinants of long-term.

Methods

From January 2005 to December 2020, we performed surgery for RSOV in 48 patients, with the mean age of 30.17±11.5 years. Because we chose to review our outcome following surgical repair, patients who were clinically diagnosed with RSOV, but refused surgery were excluded from the study. In our context, RSOV was diagnosed mostly in the third decade of life mean age or median age, range varied from 12 to 63 years at the time of surgery. Preoperative variables along with associated anomalies are listed in Table 1.

These patients were reviewed after we received approval from the institutional review committee. The preoperative characteristics along with the early and late outcomes following surgical repair were assessed retrospectively. Patients were followed up at our clinic at 2 weeks, 3 months, and then yearly following surgical repair. Transthoracic echocardiograms were done at 3 months and then yearly. Patients who were lost in outpatient clinic follow-up were telephoned and interviewed to record missing information. The last follow-up visits of the patients, in person or tele-consultation were considered as the last follow-up for those patients.

Surgical repair was carried out using cardiopulmonary bypass with moderate hypothermia through midline sternotomy. After aortic cross clamping, and aortotomy cold blood antegrade or retrograde cardioplegia (in case of significant aortic regurgitation) was given. Right atrial chamber was opened and exit site of RSOV was examined. Cardiac chamber involved was opened depending on the preference of individual surgeon, and exit site of RSOV was examined. The windsock of aneurysm was excised in all the patients and the defect was repaired primarily with valve aneurysm tissue (n1) or by patching with pericardial patch (n3) or PTFE patch (n9). Aortic end was repaired with pericardial patch, or PTFE patch. Atrial or ventricular end was repaired by primary repair using valve aneurysm tissue or by patching with pericardial patch or PTFE patch. Any coexisting defects were also repaired simultaneously.

Statistical analysis:Descriptive statistics for categorical variables are reported as frequency distribution and percentages, continuous variables as the mean with standard deviation or median and ranges, as appropriate. All analyses were performed systematically using SPSS 16 statistical software (SPSS Inc., Chicago, USA. A p-value <0.05 was considered to be statistically significant.

Results

RSOV, either congenital or acquired, is a rare cardiac anomaly even in our context. We estimated the approximate incidence of SVA as 0.22%. Out of 21,647 total open heart surgeries operated and recorded at the Centre since January 2006 to December 2020 over a period of 15 years, the surgery for RSOV comprised 0.22% (2.2 cases/1000) The frequency of RSOV was more prevalent among the male (n=35; 72.9%) than female (n=13; 27%). Patients were ranging in age from 12 to 63 years with median age of 28 years. The baseline characteristics including presentation, cusp involvement, and site of rupture, associated anomalies and techniques used for repair are given in Table 1. Most of the patients presented with shortness of breath (64,5%) and palpitation (62.5%). Asymptomatic murmur was present in two patients (4%). Rupture of the right coronary sinus into the right atrium was the most common anatomic type lesion (25/48, 52%). Preoperative aortic regurgitation (AR) equal to or greater than grade II were seen in nine and ventricular septal defects were seen in six patients. Two patients had preoperative renal failure and two were in complete heart block.

Of nine patients with significant AR in our series, three patients with severe AR required replacement of the valve with mechanical prosthesis at the initial operation. Four of other six patients had aortic valve repair with plication of the right and left coronary cusps. Another three patients with moderate AR were treated only by repairing the RSOV or VSD. Three patients had associated with at least moderate mitral regurgitation (MR), of which two patients underwent mitral valve repair . One patient with infective endocarditis underwent mitral valve replacement with mechanical prosthesis. Three patients underwent tricuspid valve repair for associated tricuspid regurgitation. One patient had associated aortic root dilatation and underwent modified Bentall's procedure, and one had right ventricular outflow tract release for right ventricular outflow tract obstruction.

Table 1: Baseline Characteristics of the patients

Characteristics		Frequency (n=48)	Percentage
Presentation	Palpitation	30	62.5%
	Shortness of Breath	31	64.5%
	Chest Pain	10	21%
Site of Rupture	Right Atrium	23	48%
	Right Ventricle	15	31%
	Left Atrium	2	4%
	Interventricular Septum	3	6%
	Right Ventricular Outflow tract obstruction	4	8%
Cusp Involvement	Right coronary cusp	25/48	52%
	Left coronary cusp	23/48	48%
Associated Anomalies	Ventricular Septal Defect	6/48	12.5%
	Aortic Regurgitation	9/48	19%
	Atrial Septal Defect	1/48	2%
	Right Ventricular tract outflow obstruction	1/48	2%

Ruptured sinus of Valsalva aneurysm was repaired primarily (n1) or by patching: pericardial (n3) or PTFE patch (n44). Repair was done through dual chamber approach in 39 and single chamber approach in nine patients.

The median postoperative intensive care unit (ICU) stay was 2 days (range, 2–7 days; Table 2). One patient had to be reexplored (1/48, 2%) in immediate postoperative period for significant mediastinal bleeding. There was one in-hospital mortality (n=1/48) after 6 days of surgery. The patient had presented preoperatively in cardiogenic shock and had preoperative renal failure. Complete heart block was present in two patients (2/48, 4%) for which transvenous permanent pacemaker was placed post operatively. Two patients developed wound infection, grade one and grade two each for which wound debridement was done and the patient recovered well. The median postoperative hospital stay was 11 days (range, 5–38 days; Table 2). Neurological complications were not seen in any of the patients.

Table 2: Early results after RSOV repair

Postoperative outcomes	
Reexploration for significant mediastinal bleeding	1 patient (2%)
In-hospital mortality	1 patient (2%)
Duration of postoperative ICU stay (median duration in days)	2 days (range, 2–7 days)
Duration of postoperative hospital stay (median duration in days)	11 days (range, 5–38 days)

Patients were followed up at our clinic at 2 weeks, 3 months, and then yearly following surgical repair. Transthoracic echocardiograms were done at 3 months and then yearly. Mean follow up of 7.07±3.93 (range, 0.83-15) years showed no recurrence. All survivors were in New York Heart Association Functional Class I or II. There was one late death in a 56 year old female patient due to severe pneumonia who had underwent repair 5 years back Three patients had moderate aortic regurgitation, one patient had moderate mitral regurgitation grade and another one patient had at least moderate tricuspid regurgitation at the latest follow-up. All of them were managed conservatively.

Discussion

Sinus of valsalva aneurysm is a rare cardiac anomaly with etiology being either congenital or acquired. The potentially fatal complication of the lesion is its rupture. Males are more affected than females. In our study 72.9% were male (n=35). Moustafa et al study showed, non-Coronary sinus (31.2%) to be the most commonly involved followed by RCC in 22.9%. In 47.9% of patients fistula was leaking into RA and in 14% into RV. There are studies in which the ruptured aneurysm drained into IVS.[7-9] In our study it was found in 6.2%.

Nearly half of the ruptured aneurysm had associated cardiac lesion. VSD and AR were the most common in study led by Moustafa.[6] But in our study only 6 patients (12.5%) had an associated VSD. This could be because the age group of our study was is beyond the usual presentation for patients with VSD. Significant AR was seen in 19% of our patients. In our series, 3 patients with severe AR required replacement of the valve at the initial operation. 4 of other 6 patients had aortic valve repair with plication of the right and left coronary cusps. Another 3 patients with moderate AR were treated only by repairing the RSOV or VSD.

RSOV may have various presentations. The most common were palpitation (62.5%) and shortness of breath (64.5%) in our study.

The repair for RSOV may be surgical or transcatheter approach.[10,11] Surgery is performed under cardiopulmonary bypass. The approaches could be through aortotomy, through the chamber when the aneurysm has ruptured into or dual chamber of approach. Some studies suggest repair through the aortotomy to be associated with higher risk of post-operative AR compared to through the draining chamber.[2,12] However some centers prefer dual chamber approach with patch repair.[13] There are studies that show no association between the approach and post-operative progression of aortic regurgitation.[14] In our center 77% of the repair was done through dual chamber approach.

The mechanism of repair could be patch closure or direct closure depending upon the size of the defect. Study by Gupta M et al, showed low residual or recurrent fistula with the patch technique.[15] Patients with direct closure of RSOV have shown to be associated with early progression of AR.[14] Thus, studies suggest avoiding direct closure technique. As we had only one patient in whom direct closure was done, co-relation of aortic regurgitation with technique of repair is hard to decide in our study. Out of 48 patients, 45(93.7%) of the patients had patch closure. Among these 93% had PTFE patch closure and 0.07% had pericardial patch repair. None of the patients had recurrence of the fistula in the follow-up period.

The operative mortality is low for surgical repair of RSOV.[13,16-18] In our study it is 2.08%. The patient was in cardiogenic shock at the time of presentation, thus increasing the surgical risk.

Limitation

The present study being a retrospective study, we have limited follow-up data to comment on the actuarial survival rates. We also did not have enough recorded data to comment on the etiology of the RSOV, hence we failed to clarify the exact etiology of this disease process in our population.

Conclusion

Repair of RSOV carries low surgical risk and has a good prognosis when prompt diagnosis and optimal surgical management is yielded with low incidence of recurrence.

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