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Research Article

Aortic Valve Repair Following Ventriculo-septal Defect (VSD) and Severe Aortic Regurgitation: Surgical Techniques, Complications and Follow up Events

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Abstract

Background

Aortic regurgitation (AR) is a common sequel resulting from prolapsed of right coronary cusp (RCC), non-coronary cusp (NCC) or both, in the repair of ventricular septal defects (VSD).

Objectives

The purpose of presenting these cases is to highlight the fact that aortic valve repair does not only end in addressing the cusp as this will result in moderate aortic regurgitation after surgery.

Results

Our first case is a two and half year old male with a large peri-membranous VSD with RCC prolapse causing co-optation failure and severe AR .The second case is a 12 year old male a large peri-membranous ventriculo-septal defect with RCC prolapsed with severe AR .The third case is a 12 year old female with a large peri-membranous VSD with a significant right coronary cusp prolapsed, severe aortic regurgitation due to non-cooptation of RCC with the remaining cusps with flow reversal in the descending aorta. The last case is a four year, 6-month-old male with a large peri-membranous VSD and severe AR from NCC and RCC prolapse. All had repair of aortic valve with mild regurgitation after several follow up.

Conclusion

Aortic valve repair resulting from prolapse of RCC and NCC in children with VSD is expedient as it reduces morbidity and mortality post-operation.

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Keywords: Ventricular septal defects; Aortic regurgitation; Coronary cusp (RCC); Non coronary cusp (NCC).

Introduction

Aortic regurgitation (AR) with ventricular septal defects (VSD) is an anatomic anomaly resulting from prolapsed of right coronary cusp (RCC), non-coronary cusp (NCC) or both and presents in 4.5-11% of cases [1].

Aortic valves prolapse with ventricular septal defect (VSD) are seen in over 5% of children [2]. This anomaly is common with Sub-arterial VSDs and it occurs about five times compared with peri membranous VSDs [2]. Aortic regurgitation resulting from underdevelopment of the aortic valvular commissure, and that from deficiency of the canal musculature are grouped into type 1 and 2 respectively [3]. There are several theories proposed as the cause of aortic prolapsed in VSDs. This ranges from deformity of the cusps, structural defects of leaflets support, weakness of the commissures and the issues arising from the Venturi effect.

Venturi effect has been documented as a single potent factor that could give rise to development of aortic regurgitation. Identifying Venturi effect as an important pathological mechanism in AR will help in screening children with VSD who are at risk of AR. This will predict both those patients in whom surgery is indicated as well as the timing of surgical intervention.

Surgical repair of the aortic valve from aortic regurgitation (AR)in children with VSD has gained ground over the last decade. Besides, aortic valve repair still poses a serious challenge especially in resource limited countries like ours. Several surgical maneuvers abound in the repair of the aortic valve. This includes direct repair of damaged leaflets, commissurotomy, re-suspension, thinning and extension of the prolapsed leaflet. However, complex repair often require more innovative and advanced techniques. The first surgical repair was done by Starr, et al in 1960 [4]. Later, Trusler, et al. [5] and Spencer, et al. [6] introduced the plication of prolapsed leaflet using pledgeted mattress suture. Chauvaud, et al. [7] and Carpentier introduced triangular valvular resection with very good outcome [8]. Transaortic repair of prolapsed RCC with pledged sutures was done by Yacoub, et al. [9].

The report of these case series is important to enable the cardiologists and cardiac surgeons have a high index of suspicion of possibilities of VSDs especially sub-pulmonic VSDs to present with aortic regurgitation and to be equipped with skills to tackle the management and understand the numerous complications that follow this anomaly. The purpose of presenting these cases is to highlight the fact that aortic valve repair does not only end in addressing the cusp as this will result in moderate aortic regurgitation after surgery. It is expedient therefore to address every aspect from the ventriculo-arterial junction to STJ in the approach of aortic valve repair as this will always give a better result of trivial regurgitation after surgery. In addition, the size of aortic annulus to be reduced is also very important as this is often neglected among adult with AR. Early identification and appropriate intervention can significantly improve the quality of life.

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Case presentation

Our first case is a 2 and half year-old male who presented with frequent cough. Examination showed pansystolic murmur 3/6 at mid sternal boarder while Echo showed large peri membranous VSD with RCC prolapse causing cooptation failure and severe AR. The second case is a 12-year-old male presented with recurrent respiratory tract infections at infancy and dyspnoea on exertion, NYC classification 2, PSM 4/6 in the mid sterna boarder. Echo showed peri membranous ventricular septal defect with RCC prolapsed with severe AR. The third case is a 12-year-old female admitted with a failure to thrive and recurrent RTIs. Examination showed pansystolic murmur at mid sternal boarder. Echo showed large peri membranous VSD with a significant right coronary cusp prolapsed, severe aortic regurgitation due to non-cooptation of RCC with the remaining cusps with flow reversal in the descending aorta. The last case is a four year, 6-month-old male who presented with recurrent respiratory tract infection and failure to thrive. Examination revealed a pan-systolic murmur at the mid sternal boarder.

Echo showed a large peri membranous VSD and severe AR from NCC and RCC prolapse. The AR jet is along the line of cooptation of the LCC with the NCC. There is also a jet across the annulus at the RCC-NCC cooptation line. Table 1-3 showed detailed events of the subjects.

Surgical procedure

Standard midline sternotomy, thymectomy and pericardiotomy were done. PDA was dissected and ligated. Cardio-pulmonary bypass was established with aorto-bicaval cannulation after systemic heparinisation. Both cavae snugged. Transverse aortotomy was done. The heart was arrested by giving delnido cardioplegia directly into both the coronary ostia.

Main pulmonary artery opened, left heart vented through left atrium. A patch closure of VSD with 6 zero proline 10 mm predigested suture was instituted. Main pulmonary artery was closed using proline 6 zero sutures. Root of aorta was dissected and released from surrounding tissues. Semi-circumferential annuloplasty was done from NCC to LCC using a strip of Teflon. RCC and NCC were suspended to their respective commissures and RCC and NCC leaflet edges were plicated using 7 zero proline 10mm pledgeted sutures. Saline test done was found to be satisfactory. Immediate intra operative trans-thoracic echo showed moderate AR.

In case two however, Bovine pericardium was then anastomosed onto annulus using 6 zero proline sutures.

In our third case, Truslers repair done on the prolapsed NCC leaflet by placating the NCC leaflet edges to the respective commissures with 6 zero 13mm pledgeted sutures. Truslers repair of RCC was done with 6 zero proline sutures. Saline test was found to be satisfactory. The right and left coronary buttons were excised from respective coronary sinuses. The aortic root was dissected and released from surrounding tissues. Annuloplasty was done with 5 zero proline sutures.

Coronary buttons were reattached to respective sinuses with 6 zero proline sutures.

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The sinu-tubular junction plasty was done using Teflon felt to buttress the end-to-end anastomosis of the transected aorta with 6 zero proline sutures.

In our fourth series, a sub-coronary channel was created below RCA and LMCA for aortic annuloplasty. A partial circumferential annuloplasty was done using a strip of Teflon placed all around the aortic root and sub-coronary below the RCA channel. A portion of the aortic annulus below the LMCA was excluded from the annuloplasty. Immediate intra-operative transthoracic echo showed trivial AR.

The mean cross clamp time, cardiopulmonary bypass time and mean stay in intensive care unit are 124 ± 16.51, 142.25 ± 28.19 mins and 6.66 ± 0.57 days respectively.

Characteristics	Frequency (%); and mean (SD)
Gender	
Male	3(75%)
Female	1(25%)
Age in years	7.75 (4.97)
CPB and cross clamping time	Mean (SD) in (mins)
Cross clamp time	124 ± 16.51
CPB time	142.25 ± 28.19
Mean stay in intensive care unit	6.66 ± 0.57 days.

Table 1: Characteristics and socio-demographic features of subjects and surgical events.

Features	No of cases	Echo report
Cough (PSM 3'6)	3	Large PM VSD, RCC prolapse and Coapt failure sAR,
Failure to thrive (NYC 2, 4'6)	4	Large PM VSD, RCC prolapse, sAR, Coap failure
Recurrent RTIs (PSM 3'6)	4	Large PM VSD, NCC and RCC prolapsed, sAR,

Table 2: Showing clinical features and summary of preoperative Echo results.

	First Follow up	2nd follow up	3rd follow up	4th follow up	5th Follow up
Case 1	Moderate AR	Mild AR	Mild AR		
Case 2	Mild AR	Mild AR	Mild AR	Mild AR	
Case 3	Mild AR	Mild AR	Mild AR		
Case 4	Mild AR	Mild AR	Mild AR	Mild AR	

Table 3: Showing follow up events of patient with respect to Aortic regurgitation.

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Discussion

Closure of VSD does not suffice in children with moderate to severe aortic regurgitation. Re-operation may be expedient in many children because of progressive aortic regurgitation [10]. If the surgery is deferred till adulthood, repair may be difficult. It is pertinent to do an intra-operative assessment of the aortic valve in all children suspected with aortic regurgitation.

Valve repair is the treatment of choice in children with aortic valve regurgitation. However, this may not be successful. The plication of prolapsed leaflet on the aortic wall has been proposed by Trusler, et al. [10] However, recurrence has been noted as a common sequel in this type of surgery. This may cause residual regurgitation [5]. Nevertheless, our reportage showed mild or no regurgitation. This remained the same after a long follow up. Besides, Kalangos, et al. [11] noted the use of thin pericardial strip before plication at free margin of the leaflet, this technique provides a balance to the stress at commissural site with reduced incompetence in a long term. Two of the patients were given same technique and good outcome was attained. This method of repair highlighted above will help in preventing the growth of leaflets in children. Long term results are reported to be satisfactory despite different results of aortic valvuloplasty in different centers [12-14]. This is also seen in our series.

The exact measurement of the effective leaflet height suspension is a very important during surgery and this determines valve competence. In this report, the release of the cross-clamp during repair determines the real test of competence.

The association of ventricular septal defect and aortic regurgitation connotes a poor prognosis, however, with current trends in open heart surgery, total correction is being more widely advocated.

Nevertheless, total repair is difficult, and a good knowledge of the structure and function of the aortic valve is crucial. We attained complete success in all the series, all presenting with mild to no aortic regurgitation at follow up.

The age of presentation of our cases is between 2 to 12 years. The exact age prevalence of AR is unknown; however, this is rare before the age of 2. Once AR develops, it worsens in severity within ten years. There may be episodes of Aneurysm of the sinuses of Valsalva in sub-pulmonic VSD.

Over two and half years, the unit has done 1000 pediatric surgical case, of which 400 were VSD closures, out of which only 4 had VSD with severe regurgitation giving a prevalence rate of 1%. This was less than that of Hasan, et al. [15] which revealed that out of 314 patients with VSD closure, 9 patients had severe AR giving a prevalence of 2.86%. Our finding is also less than 4.5-11% seen in the general population.

There is male dominance as seen in this study. This was in tandem with Hasan's study but however different from that of Francois, et al. who noted no difference in gender. The reason for this preponderance could be the general reason seen among males been more

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afflicted with congenital heart disease than females.

Postoperative course in all our cases were good with all having mild aortic regurgitation on follow up with no post operative morbidity and mortality. This finding is also noted by Hasan, et al. [15] who neither found any mortality nor atrioventricular block. There was no significant aortic regurgitation seen in this study post-operation. There were no complications from mechanical valve after a long follow up.

Regarding children with VSD who had aortic regurgitation, the route of surgery and the technique of VSD closure are crucial. Our cases were repaired by the aortotomy or via right atrial route in the first place. There were no reported residual VSD in our cases on follow-up. Aortic valve should be replaced without hesistance if aortic valve repair is not successful. Literature had shown that on a long-term follow-up, left ventricular functions and functional capacity may show significant improvement after aortic valve repair and replacement. This also akin to our series.

Echo showed that all our cases, but one was peri membranous VSD. This conflicts with earlier report that sub arterial VSDs are more liable to aortic regurgitation compared to peri membranous VSD.

Conclusion

Aortic valve repair resulting from prolapse of RCC and NCC in children with VSD is expedient as it reduces morbidity and mortality post-operation.

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