

Results of Simplified Single-Patch Repair for Complete Atrioventricular Septal Defect

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Abstract

Background: Since Wilcox's description of the simplified single-patch technique for atrioventricular septal defect (AVSD) repair in 1997, several studies have compared that technique with the two-patch technique.

Objective: To report the mid- and long-term results of the simplified single-patch technique for complete AVSD repair.

Methods: Retrospective study of 16 consecutive cases between January 2001 and December 2011. The patients' mean age was 18.31 ± 34.19 months (2 months - 11 years), and their mean weight, 7.80 ± 6.12 kg (3.77 – 25.0 kg). Six patients were males and 14 had Down syndrome. Mean follow-up duration was 54.97 ± 47.79 months.

Results: Mean cardiopulmonary bypass time was 74.63 ± 18.48 min (49 - 112 min), and mean aortic cross-clamp time, 46.44 ± 11.89 min (34 - 67 min). Two patients died during hospitalization (12.5%), both of cardiovascular causes. Three patients underwent reoperation due to left atrioventricular (AV) valve regurgitation, and two had third-degree AV block, requiring permanent pacemaker implantation. No patient had left ventricular outflow tract obstruction. The 14 surviving patients remain asymptomatic, ten of whom with mild left AV valve regurgitation (71.42%).

Conclusion: The simplified single-patch technique for complete AVSD repair proved to be feasible, providing adequate correction of the defects and favorable clinical and echocardiographic outcome in the mean 57.97-month follow-up (Arq Bras Cardiol. 2013;100(3):288-293).

Keywords: Heart Septal Defects, Atrial / mortality; Heart Septal Defects, Atrial / surgery; Thoracotomy.

Introduction

Atrioventricular septal defect (AVSD) occurs in two out of ten thousand live births, corresponding to 3% of congenital heart diseases. The basic defect is absence of the atrioventricular (AV) septum and usual presence of five or more leaflets of varied sizes in the common AV valve¹. Anatomical subgroups can be classified based on the insertion of the chordae tendineae and the morphology of the superior bridging leaflet of the common AV valve, according to the so-called Rastelli classification².

Because of anatomical and functional changes, the clinical findings are characterized by increased pulmonary blood flow already in the first weeks of life, leading to a 54% survival at the age of six months and a 15% survival at the age of two years^{3,4}. While congestive heart failure is the major cause of death in infants, occlusive pulmonary vascular disease predominates from the first year of life onwards^{1,3}.

Down syndrome can be found in approximately 75% of the patients with complete AVSD¹.

The most common surgical technique to repair AVSD involves the use of two patches (an atrial and a ventricular), respecting the integrity of the valvular leaflets. That approach replaced the first technique described, which consisted of a single AV patch, in which the leaflets were sectioned and re-sutured to the patch.

In 1997, Wilcox, with the aid of the pathologist Robert Anderson, described a simplified repair of the AVSD, in which no patch was implanted in the ventricular component, but the common AV valve was sutured to the crest of the ventricular septum, and the same suture stitches were used to fix the septal patch⁵. In 1999, Nicholson described his initial experience with that technique, being followed by Backer in 2007, Dragulescu in 2008, and Jeong in 2009⁶⁻⁹. That simplified repair approach, despite the good experiences reported, has been rarely disclosed and performed, being considered a second option by many or even ignored in some places. Currently, in-hospital mortality indices close to 5% are reported with that technique⁷.

This study aimed at reporting a case series of complete AVSD repair that uses the simplified single-patch technique, and at assessing its clinical, surgical and echocardiographic results in the mid and long run.

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Manuscript received March 21, 2012, revised manuscript August 15, 2012, accepted September 12, 2012.

DOI: 10.5935/abc.20130067

Patients and Methods

This is a retrospective study of a consecutive case series of 16 patients diagnosed with complete AVSD and operated on according to the simplified single-patch technique at the Instituto de Cardiologia do Rio Grande do Sul, from January 2001 to December 2011. This sample does not include all patients operated on due to AVSD at that institution during that period, but only those managed by one of the surgeons, who adopted that technique as preferential, after observing that its simplicity was accompanied by immediate anatomical and functional results similar to those of the cases submitted to the two-patch repair. Patients with incomplete AVSD or other severe malformations associated, such as tetralogy of Fallot, were excluded.

Of the 16 patients, six (37.5%) were males, 11 (68.8%) had been classified as Rastelli type A, two (12.5%) as Rastelli type C, and three had no classification described in their medical records. Fourteen (87.5%) patients had been diagnosed with Down syndrome. Their mean weight at the time of surgery was 7.80 ± 6.12 kg (3.77 – 25.0 kg), and their mean age, 18.31 ± 34.19 months (2 months - 11 years). On preoperative echocardiography, regurgitation in the common AV valve was severe in three (18.75%) patients, moderate in seven (43.75%), and mild in six (37.50%).

In the postoperative follow-up, the patients were assessed for the presence of residual ventricular septal defect (VSD) that required repair surgery, competence of the left AV valve, obstruction of the left ventricular outflow tract, and third-degree AV block (AVB) requiring permanent pacemaker implantation.

The descriptive analysis of categorical variables was performed by use of absolute and relative frequency distribution, and that of quantitative variables, by use of mean, standard deviation and median, when indicated.

Surgical technique

All patients were operated on through median sternotomy, with cannulation of the ascending aorta and inferior and superior vena cava, cardiopulmonary bypass, anterograde crystalloid hypothermic cardioplegia, and right oblique atriotomy, with maximum exposure of the AVSD.

The simplified single-patch technique developed by Wilcox was used with small adaptations⁵. It consisted of a direct suture of the ventricular component, with placement of U sutures, with 5-0 or 6-0 polyester, according to the case, tied on a Dacron fabric to the right side of the ventricular septum. Such sutures are placed right below the septal crest to avoid damaging the conducting tissue of the heart. Then the sutures are passed through the antero-superior and postero-inferior valvular leaflets at appropriate sites, demarcating the limits of the right and left atrial outflow tracts, thus determining the size of the right and left AV valve orifices. Previous 6.0 polypropylene sutures are placed on the zone of apposition of the left AV valve leaflets, prior to the sutures of the septal defect, anticipating the repair to be performed. After tying the ventricular stitches that close the ventricular component and fix the AV valves, the suture of the patch is completed continuously at the atrial level, with 5-0 or 6-0 polypropylene, usually leaving the coronary sinus to the right side. The AV valves are tested by instillation of cold saline solution into every ventricle, the repair of each valve being adjusted as

necessary, and the "cleft" of the anterior neoleaflet of the left AV valve being always sutured with interrupted 6-0 polypropylene stitches. In case of pulmonary hypertension, catheters of the left atrium and pulmonary artery are placed for monitoring.

This study had no external financing source. Its financial support consisted only of a scientific initiation grant of CNPq and Fapergs. This study was approved by the Committee on Ethics and Research of the institution where it was conducted.

Results

Table 1 shows the preoperative characteristics of the patients, and Table 2 shows the associated diseases.

The mean cardiopulmonary bypass and aortic cross-clamp times were 74.63 ± 18.48 min (49 – 112 min) and 46.44 ± 11.89 min (34 - 67 min), respectively.

Nine patients (56.3%) underwent, concomitantly with complete AVSD repair, closure of a patent ductus arteriosus.

In 57.97 ± 47.79 months (one day – 10.5 years) of mean postoperative follow-up, two patients (12.5%) died early, before hospital discharge. The first was a patient with severe pulmonary hypertension, who developed refractory cardiogenic shock and died at the 16th postoperative hour. The second patient required reoperation on the 16th postoperative day, because of severe left and right AV valvular regurgitation, and developed cardiogenic shock on the ninth postoperative day. The 14 surviving patients (87.5%) were discharged from hospital in good health conditions. Mean hospitalization time was 12.44 ± 12.22 days (3 - 56 days).

Table 1 – Preoperative clinical characteristics, n = 16

| Variable | Mean \pm SD |
|-------------------------|--------------------------|
| Age | 18.31 ± 34.19 months |
| Weight | 7.80 ± 6.12 kg |
| Male sex | 6 (37.5%) |
| Rastelli classification | |
| A | 11 (68.8%) |
| C | 2 (12.5%) |
| Not specified | 3 (18.8%) |

Table 2 – Associated comorbidities

| Comorbidity | n | % |
|---------------------------------|----|------|
| Down syndrome | 14 | 87.5 |
| Patent ductus arteriosus | 9 | 56.3 |
| Congestive heart failure | 9 | 56.3 |
| Pulmonary arterial hypertension | 5 | 31.3 |
| Anemia | 5 | 31.3 |
| Gastroesophageal reflux | 2 | 12.5 |
| Preoperative renal failure | 1 | 6.3 |

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Two patients (12.5%) had first-degree AVB, requiring permanent pacemaker implantation. Three patients (18.75%) developed severe valvular regurgitation during follow-up, and underwent surgery for left AV valve repair (one by use of valvular replacement, and two by use of valvuloplasty); in one patient, the reoperation was early (within the first 30 postoperative days, already cited above among those who died), and, in two patients, late (after hospital discharge or after 30 postoperative days). Except for the patient who died, the other two undergoing reoperation had a minimum regurgitation after surgery. One of such patients had, concomitantly with AV valve regurgitation, important residual VSD, which was also corrected. No patient had obstruction of the left ventricular outflow tract.

All 14 surviving patients were asymptomatic at the last clinical review, ten of whom had mild left AV valve regurgitation on echocardiography (71.42%). Table 3 shows the results of the perioperative and late postoperative period. Table 4 compares the left AV valve regurgitation degree in the pre- and postoperative periods of surviving patients. Table 5 shows the preoperative characteristics and postoperative outcomes for every 16 patients operated on.

Discussion

Atrioventricular septal defect, also called common AV canal, is characterized by septal tissue absence or deficiency immediately above and below the level of the AV valves¹.

Atrioventricular septal defect occurs in two out of ten thousand live births, corresponding to 3% of congenital heart diseases. Both sexes are affected, but AVSD is more frequent in females (female/male ratio: 1.3/1)^{2,3}.

Minor or major cardiac malformations can be associated, as well as Down syndrome, present in approximately 75% of the patients with complete AVSD¹.

Table 3 – Results after surgery, n = 16

| Event | N | % |
|--|---|------|
| Cardiovascular mortality | 2 | 12.5 |
| Third-degree AV block requiring pacemaker | 2 | 12.5 |
| Reoperation due to left AV valve regurgitation | | |
| Early | 1 | 6.25 |
| Late | 2 | 12.5 |
| Left ventricular outflow tract obstruction | 0 | 0 |
| Renal failure requiring dialysis | 2 | 12.5 |
| Sepsis | 2 | 12.5 |

AV: atrioventricular.

Table 4 – Degree of left atrioventricular valve regurgitation, n = 14 (death excluded)

| | Preoperative | Postoperative |
|----------|--------------|---------------|
| Mild | 5 (35.71%) | 10 (71.42%) |
| Moderate | 6 (42.85%) | 3 (21.42%) |
| Severe | 3 (21.42%) | 1 (7.14%) |

Table 5 – Preoperative clinical characteristics of the 16 patients and major postoperative outcomes

| Patient | Age at surgery (months) | Weight at surgery (kg) | Degree of preoperative LAVV regurgitation | Rastelli classification | Degree of postoperative LAVV regurgitation | Length of stay (days) | Outcome |
|---------|-------------------------|------------------------|---|-------------------------|--|-----------------------|-----------------------|
| 1 | 3 | 3.7 | Mild | A | Moderate | 56 | Discharge |
| 2 | 19 | 14.0 | Moderate | A | Mild | 9 | Discharge |
| 3 | 3 | 4.62 | Severe | A | Severe | 8 | Reoperation |
| 4 | 132 | 25.0 | Moderate | A | Moderate | 7 | Discharge |
| 5 | 6 | 5.0 | Moderate | A | Mild | 8 | Discharge |
| 6 | 7 | 6.0 | Severe | C | Mild | 8 | Discharge |
| 7 | 7 | 5.8 | Mild | A | - | 3 | Death |
| 8 | 10 | 6.7 | Severe | - | Mild | 9 | Reoperation |
| 9 | 8 | 5.40 | Mild | A | Mild | 7 | Discharge |
| 10 | 68 | 19.0 | Moderate | - | Mild | 8 | Discharge |
| 11 | 5 | 5.03 | Moderate | C | Mild | 21 | Discharge |
| 12 | 3 | 4.0 | Moderate | A | - | 13 | Reoperation and Death |
| 13 | 6 | 3.87 | Mild | A | Mild | 10 | Discharge |
| 14 | 6 | 6.5 | Moderate | - | Mild | 10 | Discharge |
| 15 | 8 | 5.55 | Mild | A | Moderate | 12 | Discharge |
| 16 | 2 | 4.5 | Mild | A | Mild | 10 | Discharge |

LAVV: left atrioventricular valve.

Anatomical subgroups can be classified based on the insertion of the chordae tendineae and the morphology of the superior bridging leaflet of the common AV valve. In type A, the superior bridging leaflet is almost totally attached to the left ventricle and firmly fixed over the ventricular septum by multiple insertions of the chordae. In type B, the superior bridging leaflet is larger and hangs over the ventricular septum more than in type A, attached to it by an anomalous papillary muscle of the right ventricle. In type C, the superior bridging leaflet is larger and unattached to the ventricular septum (free-floating leaflet), causing an unrestricted VSD².

The first surgical repair of complete AVSD was reported by Lillehei et al¹⁰ in 1955 and consisted of suturing the atrial border of the septal defect to the ventricular septum crest. The first experiences with complete AVSD were associated with high in-hospital mortality, frequently related to complete heart block, left AV valve regurgitation after repair or development of subaortic stenosis¹. Since then, several repair techniques for that defect have been described, involving the use of either a single patch or two patches; the results have improved as knowledge on the anatomy of the lesion increased and surgical management and postoperative care advanced. Current mortality rates close to 5% have been reported⁷.

The classic single-patch technique was described for the first time by Maloney et al¹¹ in 1962 and consists of suturing an autologous pericardial patch to the ventricular and atrial portions. In 1976, Trusler et al¹² introduced the double-patch technique with prosthetic material for the AVSD. Both techniques have resulted in adequate postoperative AV valve function; left AV valve regurgitation, however, have remained an important cause of postoperative morbidity in both techniques, with reoperation rate ranging from 6% to 15%^{3,5}.

In 1968, a new repair technique for AVSD with a single patch was described by Rastelli et al¹³. It involved the division of the common anterior and posterior leaflets with fixation to the single patch, which required the incorporation of the leaflet tissue in the suture line. Occasional dehiscence in that suture line caused valvular incompetence and was an important risk factor for reoperation and death.

More than two decades later, Merrill et al¹⁴ reported the results of surgical correction using the classic single-patch technique in 103 children, whose mean age was 6.2 months and mean weight, 5.8 kg. Those authors found a 17.47% postoperative mortality (18 deaths), and recommended that technique, whenever possible, for the definitive correction of the defect.

In 1997, Wilcox et al⁵ described a simplified AVSD correction, in which no patch was implanted in the ventricular component, but the common AV valve was sutured to the crest of the ventricular septum, by using the same suture stitches to fix the septal patch, which was placed only in the atrial component⁵. In that study, the authors have retrospectively assessed 21 patients undergoing repair of complete AVSD from January 1992 to July 1995, as follows: in nine patients, the single- or two-patch techniques, then considered standard, were used, and, in the 12 remaining patients, direct closure of the ventricular component was performed, with aortic cross-clamp and cardiopulmonary bypass times shorter, and

results, after a 34-month follow-up, similar to those of the other techniques. It is worth noting that, in that study, no patient with a very large ventricular component was included, aiming at preventing both tension in the valve leaflets and AV valve incompetence after repair.

In 1999, Nicholson et al⁶ reported 47 consecutive patients undergoing repair of complete AVSD from May 1995 to August 1998 with the use of a modified technique, which included direct suturing of the common AV valve leaflets to the crest of the ventricular septum, regardless of the size of the AVSD on preoperative echocardiography. The residual atrial septal defect (ostium primum) was closed with pericardial patch and the neomitral cleft was repaired. In that study, mortality was 4%, with a mean age at repair of 5.6 months, and 40% of the patients had associated lesions (persistent ductus arteriosus, muscle ventricular septal defect, total anomalous pulmonary venous drainage, coarctation of the aorta, and pulmonary stenosis). Neither AVB, nor residual septal defect nor left ventricular outflow tract obstruction was identified. Postoperative mitral valve incompetence was observed in 28% of the patients.

Although the single-patch technique is attractive, with shorter cardiopulmonary bypass and aortic cross-clamp times, supporters of the older techniques have emphasized its potential to obstruct the left ventricular outflow tract caused by the inferior retraction of the AV valve towards the crest of the ventricular septum, as well as its potential to cause residual VSD and to increase AV valve regurgitation due to the inferior displacement of the valve¹⁵.

Currently, the tendency is to correct AVSD at increasingly earlier ages, before the progression of pulmonary hypertension, cardiomegaly and AV valve regurgitation; thus, the simplified single-patch technique would make the surgery considerably easier in smaller children, because the lack of the ventricular patch decreases the difficulty of the procedure⁷. Other advantages of that technique as compared with the two-patch one are the better exposure and visualization of the AVSD and subvalvular apparatus, fewer sutures close to the valvular plane, and lower likelihood of damaging the heart conduction band⁵.

In 2007, Backer et al⁷ reported a cohort of 55 children with complete AVSD corrected between 2000 and 2006, as follows: 26 with the simplified single-patch technique and 29 with the classic two-patch technique. The aortic cross-clamp and cardiopulmonary bypass times were significantly shorter in the simplified technique (97.3 ± 19.9 vs. 132.3 ± 28.2 min, $p < 0.0003$; 128 ± 25 vs. 157 ± 37 min, $p < 0.03$), and there was no difference in the degree of postoperative right or left AV valve regurgitation. In addition, patients undergoing the single-patch repair showed neither third-degree AVB, nor reoperations for residual VSD nor left ventricular outflow tract obstruction. Among patients undergoing the two-patch repair, one had third-degree AVB and another had residual VSD, which required reoperation. Those authors have concluded that the simplified single-patch technique produced results comparable with the classic two-patch technique and significantly reduces the aortic cross-clamp and cardiopulmonary bypass times.

Original Article

In a review published in 2009, Backer et al¹⁶ reported their convictions that the modified single-patch technique is the best technique available to repair complete AVSD, based on the fact that its operative mortality is very low, the technique is easily applied to small children, and the incidence of reoperation due to either left AV valve regurgitation or left ventricular outflow tract obstruction is significantly lower than the previously reported¹⁶.

Regarding more recent data on mortality rate, Dragulescu et al⁸ published in 2008 their results with 107 children undergoing operation with the Rastelli single-patch procedure between 1984 and 2005. They reported an early survival of $86\% \pm 3.0\%$, and five early reoperations (two due to residual VSD and three mitral valve repair)⁸. Nine patients underwent late reoperations as follows: four for subaortic stenosis repair and five for mitral valve repair. Late survival at 10 and 15 years was $84\% \pm 3\%$.

Another important study⁹ comparing the simplified and the classic techniques for complete AVSD repair was published in 2009 with 61 patients, 18 of whom in the simplified single-patch technique group and 43 in the classic two-patch technique group. In that study, the cross-clamp time was significantly shorter in the single-patch technique group (110.8 ± 27.5 min vs. 134.4 ± 42.5 min, $p = 0.03$). During follow-up, reoperations were required as follows: two patients in the single-patch technique group because of AV valve dysfunction; and three patients in the classic two-patch technique group ($p = 0.63$). One patient died in the single-patch technique group and two died in the classic two-patch technique group ($p = 0.90$). As an advantage of the single-patch repair, those authors have emphasized, in addition to the shorter cross-clamp time, its simplicity resulting from not having to use a patch on the ventricular septum defect⁹.

Finally, a recent study¹⁷ was published in January 2012, aiming at assessing the long-term results of the reoperation for left AV valve regurgitation following AVSD repair. In that study, of the 312 patients initially undergoing repair, 45 (14.42%) required reoperation between December 1976 and July 2006, with a mortality rate of only 2% in 15 years of follow-up after primary repair for AVSD¹⁷.

Figures describing the simplified technique for complete AVSD repair can be found in the references cited, such as the study by Backer, which includes drawings showing the steps of the simplified single-patch technique, suture threads used, and results obtained⁷.

Despite the limitations related to the small number of patients, our initial experience with the simplified technique evidenced the relatively short aortic cross-clamp and cardiopulmonary bypass times, in accordance with the literature. There was no case of left ventricular outflow tract obstruction and only one case of important residual VSD, indicating that the technique is safe regarding those complications. The complications related to the left AV valve and heart block can occur and are expected with any technique. Patients with such complications were managed with reoperation for valve plasty or replacement and permanent

pacemaker implantation, respectively. It is worth noting that this study's mean follow-up was shorter than five years, including recent cases and others operated on 11 years before.

The 12.5% mortality observed resulted from immediate severe postoperative complications in two of the 16 patients undergoing surgery. The impact of one death in a small case series is always significant. However, such immediate mortality can be considered within an acceptable range, as compared with data from the international literature. Similarly, the rate of complications, such as residual VSD, third-degree AVB, and AV valve regurgitation, can also be considered within an acceptable range. It is worth noting that no late deaths were observed in this study.

Some national and international centers of excellence, conducted by surgeons exceptionally skilled, might be reporting very reduced percentages of mortality and complications, included in the lowest morbidity and mortality ranges of the registries, such as STS and EACTS. The interpretation of such data, however, should consider that those results are based on large case series originating from many institutions, unlike our case series. In the above cited bibliographic review, we found case series with 12, 47, 26 and 18 patients, in which the simplified technique was used^{5-7,9}.

Conclusion

The simplified single-patch technique for the repair of complete AVSD is feasible and showed good mid- and long-term clinical, surgical and echocardiographic results regarding both safety and survival of the 16 patients assessed. Because this is a small case series, new studies are necessary to refine the indications of the technique and compare it with other techniques available.

Author contributions

Conception and design of the research and Acquisition of data: Tagliari AP, Schröder DA, Teixeira Filho G, Prates PR, Sant'Anna JRM, Nesralla IA, Kalil RAK; Analysis and interpretation of the data and Critical revision of the manuscript for intellectual content: Tagliari AP, Kalil RAK; Statistical analysis: Tagliari AP; Writing of the manuscript: Tagliari AP, Schröder DA, Kalil RAK.

Potential Conflict of Interest

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

Sources of Funding

There were no external funding sources for this study.

Study Association

This study is not associated with any post-graduation program.

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