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Repair of Complete Atrioventricular Septal Defects with Tetralogy of Fallot, Double Patch Technique with Augmentation of the Left Atrioventricular Valve: Early Results from a Developing Country Tertiary Cardiac Centre

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Abstract

Background: The association of the Tetralogy of Fallot (TOF) with a Complete Atrioventricular Septal Defect (CAVSD) occurs in 5-10% of hearts with CAVSD. In this paper, we describe the early results of a modification of the double patch technique in which we augment the LAVV leaflet using the patch used for the VSD closure in the repair of CAVSD with TOF.

Patients and Methods: Between 2012 and 2014, 12 consecutive patients with CAVSD associated with TOF underwent complete repair were retrospectively reviewed. A double patch technique with LAVV leaflet augmentation was used in all the patients. Tetralogy of Fallot repair included transannular patch in 11 (65%) patients and valve-sparing in 6 (35%) patients.

Results: Mean age was 25 months ± 18.8, 7 males. Mean weight was 25 kg ± 18.8. Mean pulmonary artery annulus z-score was -1.2 ± 2. The mean preoperative RVOT gradient was 74 ± 18 mm Hg. The in-hospital mortality rates were 20% (2 out of 10). Mean ICU STAY time was 4.5 ± 3.5 days. Mean Mechanical ventilation time was 18 hours ± 10.3. Mean Hospital Stay was 12.5 days ± 5. Pulmonary Regurge was mild in 4 patients (40%) and moderate in the remaining six patients (60%). Postoperative (Left AV Valve Regurge) was mild to moderate in all the corrected patients. Mean follow-up was 10 ± 3.1 months (range 4 to 14 months).

Conclusion: Complete repair with this technique offer an acceptable early outcome regarding mortality, and degree of LAVV regurge.

Keywords: Complete atrio-ventricular defect; Fallot's tetralogy; Double patch technique

Abbreviations TOF: Tetralogy of Fallot; CAVSD: Complete Atrioventricular Septal Defect; LAVV: Left Atrio-Ventricular Defect; RAVV: Right Atrio-Ventricular Defect; VSD: Ventricular Septal Defect; RV: Right Ventricle; RVOT: Right Ventricular Outflow Tract; CAVC: Complete Atrio-Ventricular Canal; RVOTO: Right Ventricular Outflow Tract Obstruction; AV: Atrio-Ventricular; MV: Mitral Valve; PA: Pulmonary Artery; PV: Pulmonary Valve; LSVC: Left Superior Vena Cava; ICU: Intensive Care Unit; LVOTO: Left Ventricular Outflow Tract Obstruction

Introduction

Atrioventricular canal and Conotruncal anomalies are a heterogeneous group of lesions presenting unique challenges for surgical repair. The association of CAVSD with features of tetralogy of Fallot occurs in about 5 to 10% of the hearts with complete AVSD. 2% of tetralogy of Fallot patients present with features of septal defects [1].

The lesion complex shows the cardinal features of both constituting lesions. Attributes of atrioventricular septal defects specifically the inlet type VSD, ostium primum defect, and a common Penta-leaflet atrioventricular valve are present. Also, features of tetralogy of Fallot are also present namely the anterior deviation of the infundibular septum with overriding of the aortic annulus and the varying degrees of RV outflow obstruction [2].

Down syndrome (trisomy 21) is the most prevalent genetic syndrome associated with this lesion complex [3].

Early attempts at surgical repairs of this lesion complex were associated with high mortality. Recent series report improved outcome. Controversy exists regarding the surgical approach and techniques used in repairing various constituting lesions which varied considerably between different series. Double-patch *vs.* single-patch techniques were used for repairing the AV septal lesions. Also, RVOT obstruction was managed with transannular patch and with more conservative techniques as pulmonary valve sparing techniques. Also, the timing of complete repair is not well defined in the literature yet. There is debate regarding the use of initial palliation as a step to facilitate total repair afterward [4-15].

The aim of this report is to describe the basis of our surgical approach, to explore the results of our series in the management for CAVSD-TOF, and to consider our results in relation to previously reported series.

Patients and Methods

Patient characteristics

Between January 2012 and December 2014, 12 consecutive children with CAVSD-TOF underwent a complete correction in a single tertiary centre.

We included all patients having CAVC with TOF with suitable pulmonary anatomy and well-developed balanced ventricles.

We excluded from the study all the patients with small pulmonary arteries, pulmonary atresia, atrioventricular or ventriculoarterial discordance, unbalanced ventricles, or severe hypoplasia of either ventricle.

During the same period, 52 patients presenting CAVSD alone and 92 patients with TOF alone underwent corrective surgery in our hospital.

All patients were evaluated preoperatively using echocardiography and multislice computed tomography. Both examinations revealed a common Penta-leaflet AV valve, anterior deviation of the infundibular conal septum with more than 50% aortic overriding, and a nonrestrictive malalignment VSD with inlet extension. The Rastelli classification of the CAVSD was type B in three patients and type C in the 7 other patients. RVOTO with infundibular pulmonary stenosis was present in all patients.

Operative technique

AortoBicaval cardiopulmonary bypass with moderate systemic hypothermia (28°C) was employed in all cases. The left side of the heart was vented through the right superior pulmonary vein.

Intermittent antegrade cold Custodiol (Essential Pharmaceuticals, LLC, USA) (HTK cardioplegia) was employed in every patient for myocardial protection. Topical hypothermia was also added. Control of All preexisting shunts was done before the institution of the cardiopulmonary bypass to ensure adequate systemic perfusion. The lesion was exposed through a right oblique atriotomy in all patients. The AV anatomy was examined by saline injection testing allowing the leaflets to flow into their functional position. This is done to show the ideal site for coaptation of the superior and inferior bridging leaflets allowing cleft closure and valve repair. Marking stitches are used to keep these sites known through the procedure. We started by making incisions in the superior and inferior valve leaflets to separate the common pentaleaflet valve into two Left (LAVV) and Right Atrioventricular (RAVV) components. Care is made to provide more tissue for the LAVV reconstruction by making these incisions along the RV aspect of the ventricular septal crest. All patients underwent closure of the AV septal defect with a doublepatch technique, using two autologous pericardial patches with one fixed in a glutaraldehyde based solution. The edges of the VSD became clearly visualized and identified after release of the leaflet tissue. The fixed pericardial patch was tailored to the VSD dimensions with anterior redundancy and with the needed augmentation for the LAVV leaflets. Continuous running sutures were used to sew the patch along the ridge of the muscular septum. Visualization of the anterosuperior extension of the VSD was aided by exposure through a trans-ventricular approach. In these cases, the suture was withdrawn through the ventriculotomy, and the patch was sewn around the aortic root due to the aortic overriding. When the annulus was reached posteriorly, the suture line was carried away from the edge by 3 to 4 mm to avoid injuring the AV conduction bundle. Augmentation of the LAVV was done by using part of the pericardial patch that was deflected toward the free margin of the incised LAVV leaflets. The free edge of the LAVV was sutured to the reflected patch component using interrupted sutures passing through the patch free edge. The atrial component was closed after repairing the LAVV. A 2nd non-fixed fresh pericardial patch was used to close the ostium primum defect. Special attention is given to the relation of the conduction tissue to the margins of the ostium primum defect. We started inferolateral to the lateral side of the coronary sinus and then went to a more medial position just below the anterior lip of the coronary sinus thus avoiding injury to the conduction tissue. The coronary sinus was left to drain physiologically into the right atrium (Figures 1-4) [16-19].

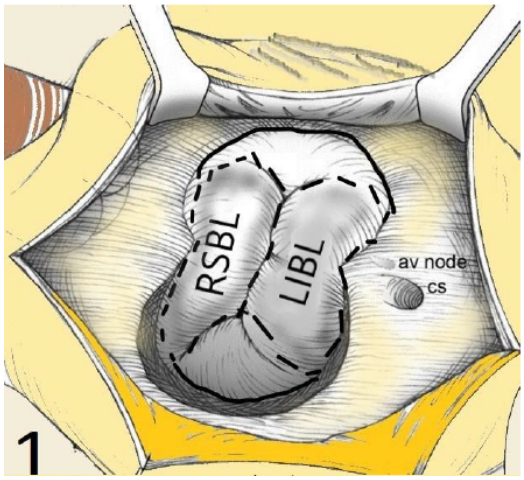

Figure 1: After right oblique atriotomy, the left and right ventricles are filled with cold saline injection solution, and the coaptation site of the superior and inferior bridging leaflets is decided.

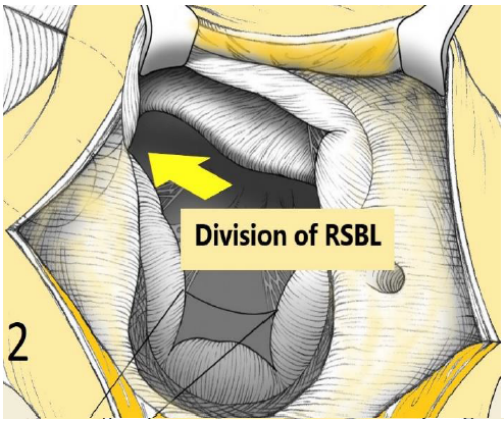

Figure 2: The superior bridging leaflet is divided and everted with retraction sutures to provide better exposure of the margins of the interventricular crest with a pilot stitch at the coaptation sites of the newly created left atrioventricular valve leaflets.

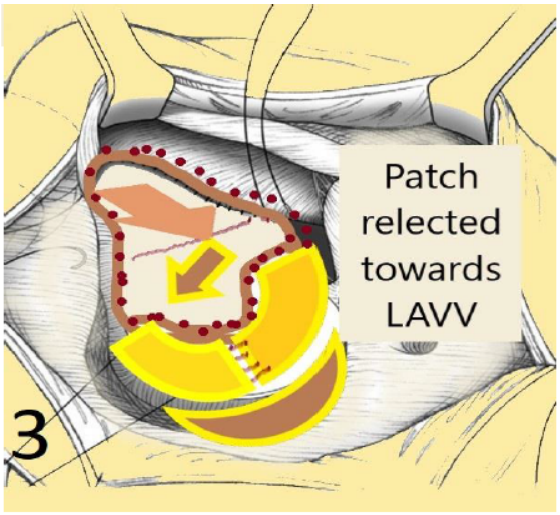

Figure 3: Modified double patch technique for the complete atrioventricular septal defect. The first patch is extended toward the left atrioventricular valve for augmentation with the free edge of the patch being sutured to the incised edge of the LAVV. The right atrioventricular is attached in the same level line with the left atrioventricular valve keeping them at the same height.

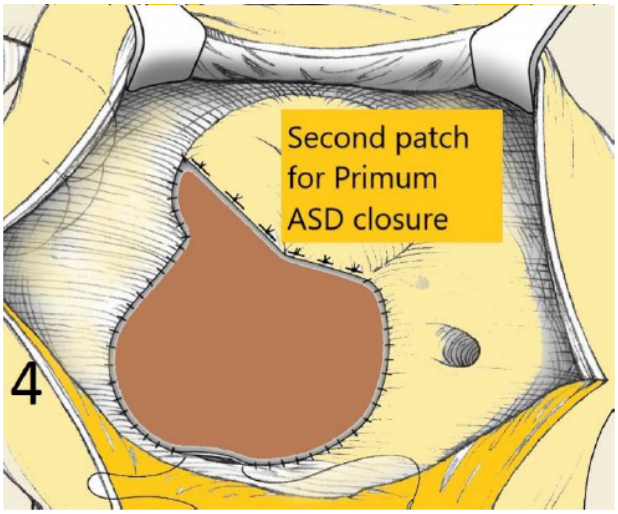

Figure 4: The second patch is sutured to the margin of the primum defect taking origin from the reflection edge of the first patch. LIBL: Left Inferior Bridging Leaflet; RSBL: Right Superior Bridging Leaflet.

After both superior and inferior bridging leaflets were sewn to the pericardial patch, the cleft in the LAVV was approximated with interrupted sutures in each patient. The competence of the LAVV was tested by closing the LV venting tube and filling the LV with a cold saline solution. LAVV annuloplasty (posterior and/or bilateral commissural annuloplasty) was performed in case of annular dilatation as a supportive procedure to the cleft closure. At the end of the corrective annuloplasty procedure, the MV annulus was sized with the proper Hegar dilators. Other procedures included resection of secondary chordae, thus increasing the mobility of the left inferior leaflet. The RAVV insufficiency was treated using commissuroplasty. A DeVega annuloplasty was employed in cases where commissuroplasty was not sufficient [20-22].

Right ventricular tract management

The main PA was opened in all patients. The PV was carefully explored, and commissurotomy was done if necessary. The size of the pulmonary annulus was determined by use of the expected Hegar dilator. Resection of the RVOT was done through the pulmonary valve, and through the tricuspid valve. In certain cases, we used a right infundibular incision-limited ventriculotomy incision-in cases of marked annular hypoplasia and severe RVOT obstruction. The transannular repair was performed when the PV annulus was more than 1 mm to 2 mm smaller than the expected hegar size estimated using Z-scores analysis. Annular sparing techniques were used otherwise. Sometimes a separate infundibular incision is used to aid in the exposure of the VSD or in cases of a severe degree of overriding for fear of residual RVOTO [22-24].

Associated surgical procedures

The associated cardiac malformations were also corrected during the same surgical procedure. In one patient the anomalous left SVC connected to the leftsided atrium was repaired by rerouting the LSVC connection using a separate pericardial patch to the right atrium. Inter-atrial ostium secundum defect septal defect was closed using a continuous suture or with integration in the patch of the ostium primum.

Statistical analysis

Descriptive statistics were used to organize data. Categorical data were presented as proportions, and continuous data were expressed as mean ± SD.

Results

Preoperative results

The preoperative findings and demographic characteristics are given in Tables 1 and 2.

Variables	No. (%)
Mean age at repair (months)	25 ± 18.8
Mean weight at repair (kg)	14.6 ± 5.37
Previous palliation (MBTS Lt. Thoracotomy)	1 (10%)
Mean age at palliation (months)	2 months
Down syndrome	6 (60%)
Pulmonary Z-score	-1.2 ± 2
Associated anomalies	
Ostium Secundum	5 (50%)
LSVC	2 (20%)

Mean gradient across the right ventricular outflow tract (mmHg)	74 ± 12
Mean arterial oxygen saturation (%)	82 ± 16
Rastelli classification	
Type A	—
Type B	3 (30%)
Type C	7 (70%)
Aorta overriding >50%	9 (90%)

Table 1: Preoperative demographic and clinical data.

Patient No.	Sex	Age (months)	Weight (kg)	Genetic syndrome	Correction (RVOT)	Early outcome
1	M	18	12	-----	Valve sparing	Alive
2	F	20	14	Down's syndrome	Transannular patch	Death
3	M	12	10.8	Down's syndrome	Valve sparing	Alive
4	M	11	8.5	Down's syndrome	Infundibular patch	Alive
5	M	10	10	-----	Infundibular patch	Alive
6	M	60	22	-----	Infundibular patch	Alive
7	M	18	13	-----	Infundibular patch	Alive
8	M	20	13	Down's syndrome	Infundibular patch	Death
9	F	60	25	Down's syndrome	Infundibular patch	Alive
10	F	22	18	Down's syndrome	Infundibular patch	Alive

Table 2: Summary of patient data.

Operative details

Complete repair was possible in all patients. The mean aortic cross-clamping time was 120 ± 25 minutes and the mean cardiopulmonary bypass time was 180 ± 25 minutes. We had 2 (20%) in-hospital mortalities in this series. These patients presented with preoperative congestive heart failure. Causes of death included progressive heart failure in patient 1 and sepsis and multiple organ failures in patient 8 (Table 3). During the same hospitalization, patient 8 was re-explored for significant bleeding and tamponade. This patient died 7 days after re-operation due to sepsis. No patients developed Renal failure needing peritoneal dialysis or ultrafiltration. Dysrhythmias (JET: Junctional Ectopic Tachycardia) were identified in 2 patients out of the 10 patients (20%). These patients had repairs requiring a transannular patch. One patient developed post-operative generalized convulsions. The mean intensive care unit stay was 4.5 ± 3.5 days. Mean duration of the mechanical ventilation was 18 hours ± 10.3. Mean hospital stay was 12.5 days ± 5.

Variables	**No. (%)**
Cardiopulmonary bypass time (minutes)	180 ± 25
Aortic cross-clamping time (minutes)	120 ± 25
Left atrioventricular valve annuloplasty	8 (80%)
Right atrioventricular valve annuloplasty	3 (30%)
Transannular patch	4 (40%)
Right Ventriculotomy	8 (80%)
Valve sparing	2(20%)
Pulmonary artery branch enlargement	2 (20%)
Pulmonary Monocusp	1 (10%)
Complications	
Neurological deficit	1 (10%)
re-exploration	1 (10%)
Pneumonia	1 (10%)
Renal failure	0 (0)
Ventricular dysrhythmias	2 (20%)
Mean intensive care unit stay(days)	4.5 ± 3.5
Postoperative left atrioventricular valve LAVV regurgitation grade	1.7 ± 0.5
Residual RVOTO gradient	21.5 mmHg ±18.2
Pulmonary Regurge	
Mild	4(40%)
Moderate	6 60%
ICU stay (days)	4.5 ± 3.5
Mechanical ventilation (hours)	18 hours ± 10.3
Hospital stay (days)	12.5 days ± 5

Table 3: Intraoperative and postoperative data.

Postoperative physiology and functional status at early follow - up

The mean follow-up time was 10 ± 3.1 months (range 4 to 14 months). All 8 survivors were in NYHA functional class I or II. All patients who underwent a transannular patch repair presented moderate PV insufficiency. Most patients had moderate pulmonary regurge in the first-month echo examination. The mean gradient across the right ventricular outflow tract was 21.5 mmHg ± 18.2, significantly lower than the pre-operative gradient ($p<0.001$). At followup, all survivors had a mild to moderate LAVV regurgitation grade I or II. The LAVV regurgitation grade at follow-up was significantly lower than early postoperatively, 1.7 ± 0.5 *versus* 2.5 ± 0.8 ($p=0.002$). None of the patients developed significant LVOTO following surgery.

Discussion

Total correction in CAVSD-TOF represents a challenging surgery due to its rare incidence and specific anatomical characteristics. The unique anatomical situation of the CAVC-TOF must be carefully considered when planning surgical correction in this pathological entity.

In this pathological entity, the antro-superior border extends more anteriorly giving the VSD a subaortic extension. There is un-wedging of the aorta in the CAVSD with the aortic valve sitting anterior and above the base of the anterior bridging leaflet. This will cause the characteristic gooseneck-long LVOT appearance in RV angiography. Additionally, this configuration will also obscure the transatrial view of the VSD. In the series of Ricci et al. [18], the aorta was dextroposed in all patients especially with CAVSD Rastelli type C. In the presence of TOF will lead to even more dextroposition of the aorta, so there is more than 50% overriding of the ventricular septum. This will give a DORV like appearance in such patients.

The Rastelli type C configuration is commonly found in patients with CAVSD-TOF. In this type, the anterior bridging leaflet presents as "free-floating". This means that it is unattached to the ventricular septal crest and will extend far into the RV. However, recent anatomic studies showed that in some cases with Rastelli type C, a single thick chordae tendinea might be anchoring the anterior bridging leaflet to the interventricular crest [13,17,25].

Surgeons should allow Maximal tissue preservation for LAVV reconstruction by making the incision in the bridging leaflets more to the right aspect of the crest. In the cases where the anterior bridging leaflet is anchored to the ventricular septal crest, the surgeon must decide which chordae needs to be preserved and which needs to be sacrificed. The aim is to preserve the function and the structure of the LAVV leaflet tissue as it is the systemic AV valve later on [19].

In some neonates, cases of double orifice LAVV, and dysplastic AV tissue there might be a deficiency of leaflet tissue. Thus, attempting a repair in these situations will produce a small valve or reduction of the mobile valve area and leaflet fixation. Also, it might lead to incorporation of more leaflet tissue into the suture line. Thus, the sutures will be placed under more tension thus leading to a high rate of suture dehiscence. Altogether, this will eventually contribute to significant post-operative LAVV regurgitation. One of the solutions proposed is the augmentation of the LAVV leaflet tissue as proposed in our technique. This technical modification will provide enough additional tissue for reconstructing the LAVV appropriately in these cases with leaflet tissue deficiency. Additional benefits include increased coaptation surface, improved leaflet mobility, and tension-free closure of the cleft.

Surgical closure of the VSD in the patients with TOF-CAVD might lead to LVOTO. The inserted ventricular patch, which constitutes the right-hand wall of the LVOT, may create a narrow outflow tract and consequently obstruction when the patch is too small anteriorly. Preoperatively in the unrepaired defects especially Rastelli type C configuration and during systole; the superior bridging leaflet of the common pentaleaflet AV valve is pushed toward the atrium, and the left ventricular outflow tract opens thus is widely open [20].

Two technical points need to be considered to prevent this complication, first, the division of the bridging leaflet and the closure of the subaortic component of the VSD as much rightward as possible. This will not only ensure allocation of more tissue for the LAVV reconstruction but also create a larger LVOT.

Second, the patch should be sized appropriately to allow it to be redundant up in the superior aspect of the VSD and enlarged anteriorly. This will increase the subaortic space, avoiding LVOTO. Some surgeons open the aortic root and pass Hegar sizer through the LVOT to avoid iatrogenic LVOTO.

Due to the nature of the TOF repair in this lesion complex, PV insufficiency or residual RVOTO might occur. These post-surgical consequences might compromise the RV function by volume or pressure overload. This in effect might worsen the RAVV regurgitation. Subsequently, this might impact the short and long-term outcome of total repair. Similarly, significant residual regurgitation of the LAVV can elevate the Pulmonary Artery (PA) pressure, and this might worsen the degree of insufficiency especially if a transannular patch is used [17]. Our techniques limit these drawbacks by ensuring the maximal degree of valvular competence through leaflet augmentation, adequate resection of the RVOT and Pulmonary valve preservation .

One of the reasons for post-operative RV failure following total repair for TOF-CAVSD is a small RV cavity. Thus, it is a pre-operative requisite to evaluate how developed is the RV cavity size. If the RV is not well developed, univentricular repair techniques should be employed including one and a half repair which incorporates CAVSD-TOF repair plus bidirectional Glenn shunt.

Most children with CAVSD-TOF have symptoms delayed beyond the neonatal or even the infant period. The age of presentation depends on the degree of RVOTO in this lesion complex. Such obstruction is functionally sufficient as a PA band, protecting the pulmonary vasculature from the high pulmonary blood flow characteristic of CAVSD, thereby delaying the pulmonary venous congestion and allowing repair at a later stage. However, patients might present with Heart failure in cases of severe RVOTO and severe AV regurgitation, and this is an indication of a complete repair even at an early age. Severe cyanosis in the neonatal period indicates severe RVOTO [26].

The classic treatment for CAVSD-TOF included palliation as a first procedure which was later followed by complete repair. The arguments for staged repair included easier repair and AV reconstruction. Also, operating at a later age allows bigger valved conduits to be inserted if needed. The problem with early palliation is that it carries potential complications, including prolonged cyanosis, excessive ventricular hypertrophy. The ventricles are subjected to chronic volume overloading and thus more progression of the AV regurgitation [14].

The recent indications for palliation in the form of an MBTS are patients with small pulmonary arteries and symptomatic cyanotic neonates. In other cases where the indication for palliation is not present, complete primary repair is done possibly after the first 6 months of life.

Success after primary repair depends on multiple factors such as the age at presentation, the severity of cyanosis, pulmonary valve annulus size, size of pulmonary arteries as estimated by the Mcgoon ratio, and the presence of associated coronary anomalies such as an anomalous coronary artery crossing the RVOT [27].

D'Allaines et al. [28] in the year 1969 published the first article reporting successful correction for this malformation. Since then, more than 40 articles described complete surgical repair of CAVSD-TOF in 400 patients.

In recent series of patients with CAVSD-TOF, post-operative mortality after complete correction reduced considerably during the last decade in comparison to the previous series. Better diagnostic techniques improved perioperative care, better surgical approaches, and an improved learning curve contributed substantially to the improvement of postoperative results [26].

Post-operative factors that are related to increased mortality after repair include residual LAVV insufficiency, residual VSDs or ASDs, and residual RVOTO or LVOTO. Recent surgical techniques include approaches to minimize the occurrence of these problems.

In our series, we have employed a modified double patch technique for CAVSD-TOF in association with augmentation of the LAVV and complete or partial cleft's closure.

The proper management of the RVOT is still a topic of controversy. There are two approaches to manage the RVOTO in this lesion complex. The first is the trans-ventricular approach. An RV incision is made which can be limited to the infundibulum or might be extended to the pulmonary annulus in case of transannular patching. Transannular patching should be reserved for patients with the small or hypoplastic annulus. Advantages of this approach include better exposure of the VSD especially its anterior exposure, also, better resection and visualization of the obstructing muscle bundles. Drawbacks of this approach include post-operative RV dysfunction and subsequent arrhythmias, pulmonary valve insufficiency induced by transannular patching. Post correction pulmonary insufficiency leads to chronic volume overloading of the RV and gradual dilation and RV dysfunction especially in cases of residual LAVV regurgitation [28].

The second approach is the trans-atrial transpulmonary approach. In this technique, the VSD is closed, and the RVOT is resected without the need for RV ventriculotomy, thus, avoiding the drawbacks of ventriculotomy. This technique was advocated in the series by Karl et al. [13].

Recently a third approach is advocated using a valved conduit thus benefitting from the exposure advantage and easier access to the RVOT and at the same time avoiding the disadvantages of pulmonary insufficiency. Recently, transannular patching in the repair of TOFCAVSD was not found to be detrimental to the patient outcomes results in the short or long term [17,29].

Conclusion

CAVSD with TOF is a population of patients at a higher risk than the general CAVSD or TOF populations. Total repair can be attempted in these patients with an excellent safety margin. The policy of staged repair and early palliation might be reserved for patients with small pulmonary arteries or in patients who become symptomatic before the age of 6 months.

Our method for CAVSD-TOF repair permits good visualization of the entire VSD and exact anatomic repair has so far been associated with a good functional early outcome in all survivors. The preservation of the LAVV competency is an essential factor affecting post-operative functional status, reoperation rate, and mortality. Augmentation is a safe and secure technique that can deal with a variety of valve morphologies and limits the residual post correction LAVV regurge.

The RVOT should be managed in the same fashion as for isolated TOF; every effort should be made to minimize the RV ventriculotomy and to preserve the pulmonary valve function.

Patient characteristics were summarized using number (percent) for categorical variables and median (range) for continuous variables. Primary outcome variables were time from definitive repair to death and time from definitive repair to reoperation. Patients not experiencing an outcome were censored at their time of last follow-up. Relationships between patient characteristics and outcome variables were examined using Kaplan-Meier survival curves; comparisons were performed using the log-rank test.

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