Atrioventricular septal defect and tetralogy of Fallot – A single tertiary center experience: A retrospective review

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ABSTRACT

Background : **Complete atrioventricular septal defect (CAVSD) in association with tetralogy of Fallot is a rare and complex disease that makes its repair more difficult than repair of either lesion alone. We reviewed retrospectively our experience in managing this lesion.**

Patients and Methods : **Between February 2006 and May 2017, 16 patients who underwent repair of CAVSD/tetralogy of Fallot (TOF) were reviewed retrospectively. Fifteen patients had trisomy 21. Five patients underwent primary repair while eleven patients went for staged repair in the form of right ventricular outflow tract (RVOT) stenting (*n* = 9) or systemic to pulmonary (S-P) surgical shunt (*n* = 2). RVOT stenting has replaced surgical shunt since 2012 in our center. Early presentation with cyanosis was the main determinant factor for staged versus primary repair.**

Results : **The median age at first palliation was 46 days (range 15–99 days). The median age at total repair for both groups was 6 months (range 3–18 months); the median age for the palliated patients was 6.5 months (range 5–18 months) while the median age for primary repaired patients was 5 months (range 3–11 months). The median weight at final repair was 6.9 kg (3.7–8.2 kg). The pulmonary valve was preserved in five patients (31%), four of them had no prior palliation. Chylothorax occurred in 50% of the patients. One late mortality occurred after final repair due to sepsis.**

Conclusion : **CAVSD/TOF can be repaired with low mortality and morbidity. The use of RVOT stent has replaced the surgical (S-P) shunt with acceptable results in our center.**

Keywords : Atrioventricular valve regurgitation, Blalock–Taussig, complete atrioventricular septal defect, left ventricular outflow tract obstruction, right ventricular outflow tract

INTRODUCTION

Complete atrioventricular septal defect (CAVSD) with tetralogy of Fallot (TOF) is a complex and uncommon disease.[1-3] This disease must be considered carefully when planning surgery.

Controversies exist in the approach of repair. Some believe that primary repair is considered superior to staged repair[3,4] while others adopted staged repair with right ventricular (RV) outflow stenting or systemic to pulmonary (S-P) shunting followed by a final repair.

In this study, we present our results in managing this complex disease.

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PATIENTS AND METHODS

Between February 2006 and May 2017, we retrospectively reviewed all patients who underwent CAVSD/TOF repair in Our Lady's Children's Hospital, Crumlin, Ireland. Patients' data were collected from the patient charts, operative reports, intensive care computer-based records, echocardiography reports, discharge reports, and follow-up reports. The ethical committee approved the study.

Surgical management

Patients were managed conservatively unless they presented with severe cyanosis defined as <70% systemic saturation in our hospital protocol, where either a S-P shunt is constructed (n = 2) or lately since 2012 an RV outflow tract (RVOT) stent is inserted in the cath lab (n = 9). Hypothermic cardiopulmonary bypass (CPB) was used in all cohorts with the use of blood cardioplegia in all. All patients underwent two-patch technique to repair the CAVSD. The superior bridging leaflet was cut toward the annulus in seven patients (43%). The repair of the RVOT was achieved by transannular patch in eleven patients. Monocusp was constructed in five patients. The monocusp was constructed by autologous pericardium in two patients and by taking one leaflet with its sinus from the bovine jugular vein (Contegra®) conduit in three patients. The atrial septal defect was closed by an autologous pericardial patch in all patients. Fenestration of the interatrial septum of 4–6 mm was created in all but one patient. The decision was based on surgeon preference. Conventional ultrafiltration and modified ultrafiltration were used at the end of all cases.

Statistical analysis

Patients' data were evaluated using numbers (percentage) for categorical variables and median (range) for continuous variables.

RESULTS

A total of 16 patients underwent complete CAVSD/TOF repair. 38% (n = 6) were males and 62% (n = 10) were females. Fifteen patients (94%) have trisomy 21. Eleven patients (69%) presented early in life with significant cyanosis defined in our hospital as systemic saturation of <70%. RVOT stent was used for palliation in nine of them (56% of our cohort) and a S-P shunt in the form of modified Blalock-Taussig (BT) shunt in two patients [Figure 1]. The median age at first palliation was 46 days (range 15–99 days). The median age at total repair for both groups was 6 months (range 3–18 months); the median group for the palliated patients was 6.5 months (range 5–18 months) while the median age for primary repaired patients was 5 months (range 3–11 months). The median weight was 6.9 kg (range 3.7–8.2 kg). Five patients presented with congestive heart failure and needed a medical management before going for a primary repair. The median follow-up time was 20.5 months (range 5–130 months).

The mean CPB time was 206.5 min (range 76–350 min), and aortic cross-clamp time was 162.5 min (range 41–201 min). Nevertheless, there is no statistically significant correlation between the bypass time and cross-clamp time when we compared them to both staged and primary repair groups [Table 1]. This is probably due to the small sample size.

No early death has occurred after final repair. However, only one late death occurred about three years after final repair from meningococcal sepsis. Fifty percent of patients (n = 8) developed chylothorax, of whom, only one had pulmonary valve preservation while all the others had a transpulmonary patch. Six (75%) of these patients, who developed chylothorax, were palliated by RVOT stent (n = 5) or BT shunt (n = 1). Complete heart block which required permanent pacemaker had occurred in two patients (12.5%). Both needed a delayed sternal closure due to prolonged bypass time due to difficult exposure. Extracorporeal life support was needed in one of these patients. This patient will be discussed below.

Four patients out of nine (44%) who underwent palliation before the complete repair needed reintervention in the form of restenting or ballooning of the previous stent before the complete repair. The median time of reintervention was 2.5 months (range 2–4 months). Only one patient needed early reintervention after the complete repair. This patient is described in detail in the discussion section.

Two patients needed late reintervention after the complete repair. One patient had RVOT stent due to high RV pressure almost 1 year after the complete repair. This patient had a transannular patch with monocusp reconstruction at the time of final repair. The other patient had right and left pulmonary artery (PA) stenting 8 months postoperatively. A year later, she went for percutaneous pulmonary valve replacement [Table 2].

DISCUSSION

Repair of the atrioventricular septal defect (AVSD)/TOF comprises a real challenge for the cardiac surgeon. The repair of TOF often results in pulmonary valve insufficiency or residual stenosis. The resulting volume overload or RV hypertension can potentially worsen right atrioventricular valve (AVV) regurgitation and impact significantly on short- and long-term outcome. Significant regurgitation of the left AVV can elevate PA pressures and increase pulmonary valve insufficiency, particularly in those following transannular patch reconstruction. These factors, in combination with technical challenges, result

Table 1: Comparison between children with and without palliation at final repair

	Patients	Two-stage repair palliation	Primary repair	*P*
Preoperative				
Age (months)	6	6.5	5	0.344
Weight (kg)	6.9	6.8	7.1	0.188
Trisomy 21	15	10	5	0.519
Operative				
Valve sparing	5	1	4	0.002
Bypass	197.64	205.81	167.66	0.436
X clamp	137.07	150.45	116	0.299
ECMO	1	1	0	0.519
Length of stay				
ICU stay (days)	16.5	19.5	3.5	0.306
Hospital stay (days)	36	44	16	0.250
Complications				
Reoperation				
Early (PPM, PVR + MVR)	3	3	0	0.221
Late (RVOT stent)	1	1		0.519
Chylothorax (%)	8 (50)	6 (75)	2 (25)	0.619
Tachyarrhythmia	1	0	1	0.519
Bradyarrhythmia	2			0.341
Sepsis	5	3	2	0.639
Stroke	1	0	1	0.413
LAVVR (moderate to severe)	2	2	0	0.341
Ventricular dysfunction	1	1	0	0.519
Mortality	1 (late)	1	0	0.519

ECMO: Extracorporeal membrane oxygenator, ICU: Intensive care unit, PPM: Permanent pacemaker, PVR: Pulmonary valve replacement, MVR: Mitral valve replacement, RVOT: Right ventricular outflow tract, LAVVR: Left atrioventricular valve regurgitation

in a greater risk for repair of CAVSD/TOF than the risk for repair of either lesion alone.[5]

The degree of RVOT obstruction determines the time of intervention in these patients. When these patients present early in the neonatal period or in infancy with cyanosis, they become a real challenge to the physicians and surgeon [Figure 2]. On the other hand, RVOT obstruction protects the pulmonary vasculature and hence allows repair at an older age.

Historical recommendations for age of repair have been in the range of 4–6 years; palliation was performed in severely cyanosed patients to allow them to reach that age.[1,2,6] However, palliation with later repair carries multiple potential complications including prolonged cyanosis, excessive ventricular hypertrophy, volume loading of the ventricle, and onset of progression of AVV regurgitation.[3] Najm *et al.* believe that repair at a younger age is more favorable as demonstrated by faster hospital recovery and lower incidence of reoperation for an equivalent duration of follow-up. The patients in their cohort presented late, their youngest patient was 2 months of age and an average age of 20 months, (range 2-89 months). Our patients presented early with cyanosis median age of 46 days (range 15–99 days). We speculate that preliminary RVOT stent followed by total repair may be the procedure of choice.

Staged repair has been recommended because of high mortality rates reported in early series, especially in younger patients.[2,6] One reason proposed for delaying the complete repair beyond infancy is the belief that the leaflets of the AVV in older patients are thicker and more amenable to repair.[7] Nevertheless, some reviews of repair of AVSD in early infancy showed no relationship between age at repair and postoperative atrioventricular valve function.[8] The second reason is early presentation with small PA size. Barron *et al.*[9] reviewed the effect of RVOT stenting on subsequent surgical intervention with attention to growth of the pulmonary arteries in patients with isolated TOF or with associated lesions. The indications for RVOT stenting in their study: first, low birth weight patients with TOF presenting with cyanosis or small PA branches and second, patients with complex anatomy, as AVSD, or comorbidities that increase the risk of primary repair. They also discussed the complications of RVOT stenting including stent migration, failure of deployment, tamponade, need for reintervention, arrhythmias, stent endocarditis, coronary artery compression, difficulty of stent removal during final repair, and death. Yet they concluded that primary RVOT stenting is a valuable and safe mode of staged palliation in TOF in small neonates and children with complex anatomies and significant comorbidities and has only few complications though high incidence of transannular patch use. Four patients out of 13 of their cohort had the diagnosis of CAVSD/TOF. In our cohort, four patients required early reintervention in the form of restenting or ballooning of the previous RVOT stent. Two patients needed late restenting. One had RVOT stent due to high RV pressure while the other had branch PA stenosis. One year later, the patient had pulmonary valve replacement. Apart from one patient who developed junctional ectopic tachycardia which was managed conservatively, no other major morbidity

Table 2: Summary of patient's data

Number	Presenting symptoms	Initial palliation	Age (days)	Trisomy 21	Total repair tech	Age (months)	Complications	Postoperative echo on last follow-up (ventricular function, left, right AVVR, RVOT obstruction, and pulmonary regurgitation)	Results	Follow-up months	Reintervention postinitial palliation	Reoperation after total repair
1	Cyanosis	RVOT stent	15	Yes	TA, 2 patch, DSC	6	CHB needed PPM	Good ventricular function, mild left and right AVVR	Alive, well	5	RVOT restent	
2	Cyanosis	RVOT stent	62	Yes	PVp, 2 patch	6	JET	Good biventricular function Mild RAVVR - Mild LAVVR RVOT PG 33 mmHg Moderate PR	Alive, well	8	Balloon dilation	
3	Cyanosis	RVOT stent	46	No	TA, 2 patch, monocusp contegra, DSC	8	Needed ECMO 12 h in ICU, then had myectomy for LVOTO, pulmonary melody valve, then melody valve in LAVV due to severe left AVVR, chylothorax long ICU stay	Good biventricular function Mild+RAVVR - Mild melody valve stenosis and regurgitation Melody in RVOT PG 15 mmHg Mild-moderate PR	Alive, mild mitral melody valve stenosis and regurgitation, pulmonary melody moderate regurgitation	11	Balloon dilation	PVR, MVR
4	Cyanosis	RVOT stent	46	Yes	TA, 2 patch, monocusp contega	7	Chylothorax, long ICU stay (sepsis)	Good biventricular function Mild RAVVR - Mild LAVVR RVOT PG 25 mmHg Free PR	Alive, well	12	Balloon dilation	
5				Yes	TA, 2 patch	13		Good biventricular function Mild RAVVRv - mild LAVVR RVOT PG 25 mmHg Free PR	Alive, well	6		
6	Cyanosis	RVOT stent	29	Yes	TA, 2 patch, divided superior bridging leaflet	11	Chylothorax	Good biventricular function No RAVVR - Mild LAVVR RVOT PG 22 mmHg Free PR	Alive, well	22		
7				Yes	TA, 2 patch, monocusp pericardial	4		Good biventricular function No RAVVR - Mild LAVVR RVOT PG 45 mmHg Moderate PR	Alive, well	19		
8	Cyanosis	RVOT stent	46	Yes	PVp, 2 patch	8		Good biventricular function Mild RAVVR - Mild LAVVR RVOT PG 19 mmHg Free PR	Alive and well	10		

Contd...

Table 2: Contd...

Number	Presenting symptoms	Initial palliation	Age (days)	Trisomy 21	Total repair tech	Age (months)	Complications	Postoperative echo on last follow-up (ventricular function, left, right AVVR, RVOT obstruction, and pulmonary regurgitation)	Results	Follow-up months	Reintervention postinitial palliation	Reoperation after total repair
9	Cyanosis	RVOT stent	41	Yes	TA, 2 patch, monocusp contgra, divided superior bridging leaflet	6	Chylothorax	Good biventricular function Mild RAVVR - Mild LAVVR RVOT PG 24 mmHg moderate PR	Alive, needed RVOT stent 1 year later	32		RVOT stent
10	Cyanosis	RVOT stent	62	Yes	TA, 2 patch, monocusp perocardial	5	Chylothorax	Good biventricular function Mild RAVVR - Mild LAVVR RVOT PG 16 mmHg Free PR	Alive, well	30		
11				Yes	TA, 2 patch	5	Chylothorax, long ICU staym, CHB needed PPM (sepsis)	Good biventricular function Trivial RAVVR - Mild LAVVR RVOT PG 25 mmHg Mild PR	Alive, well	39		
12				Yes	PVp, 2 patch	5		Good biventricular function Trivial RAVVR - Mild LAVVR RVOT PG 25 mmHg Mild PR	Alive, well	79		
13	Cyanosis	BT shunt	98	Yes	PVp, 2 patch	4		Good biventricular function Mild RAVVR - Mild LAVVR RVOT PG 14 mmHg Free PR	Alive, well	88		
14	Cyanosis	BT shunt	18	Yes	TA, 2 patch	16	Chylothorax	Good biventricular function Mild RAVVR - Moderate LAVVR RVOT PG 15 mmHg Free PR	Alive, well	11		
15				Yes	TA, 2 patch	18		Good biventricular function Mild RAVVR - Mild LAVVR RVOT PG 15 mmHg Free PR	Died 3 years later due to sepsis	24		
16				Yes	PVp, 2 patch	3	Chylothorax	Good biventricular function Trivial RAVVR – Mild LAVVR RVOT PG 25 mmHg Mild PR	Alive, well	130		

CHB: Complete heart block, BT: Blalock–Taussig, DSC: Delayed sternal closure, ICU: Intensive care unit, JET: Junctional ectopic tachycardia, LAVV: Left atrioventricular valve, LVOTO: Left ventricular outflow tract obstruction, MVR: Mitral valve replacement, PPM: Permanent pacemaker, PVp: Pulmonary valve preservation, PVR: Pulmonary valve replacement, RVOT: Right ventricular outflow tract, TA: Transannular patch, ECMO: Extracorporeal membrane oxygenator, AVVR: Atrioventricular valve regurgitation, LAVVR: Left AVVR, RAVVR: Right AVVR, CHF : Congestive heart failure, MBT : Modified blalock taussig shunt, PG: Pressure gradient, PR: Pulmonary regurgitation

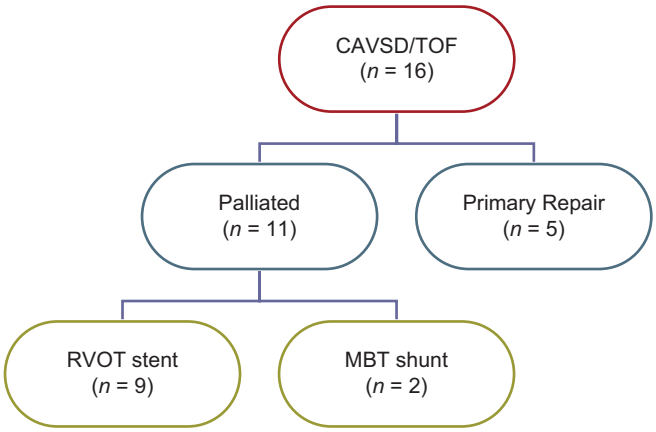

Figure 1: Flow chart demonstrating the number of patients who underwent palliation and primary repair and type of palliation

or death was observed. Explanting the stent during the final repair in our series, as in theirs, did not make the placement of the ventricular septal defect sutures difficult.

There is increasing enthusiasm for single-stage repair for patients with the diagnosis of TOF only, even in symptomatic neonates; but in most cases, the outcomes are strikingly different from those achieved in older age groups. The Southern Thoracic Society series analyzed over 3000 TOF repairs (2002–2007) and showed a 7.3% mortality compared with 1.5% in older infants.[9] The operative mortality and morbidity associated with the surgical BT shunts drove the enthusiasm toward primary repair.[10] These concerns have led to an increasing popularity of primary RVOT stenting as an alternative to the conventional staged approach using S-P shunting in such lesions.[9]

The decision to send our palliated patients to surgery was made based on close follow-up by the cardiologist. Once the patient reached our target age (4–6 months) or target weight (4–6 kg), the cardiologist rediscusses the case in our combined cardiology and cardiac surgery meeting and sends the patient for surgery.

Left ventricular outflow obstruction was encountered in only one case in our series. This patient was a 7-month-old boy with history of neonatal palliative RVOT stenting. He had a preoperative significant common AVV regurgitation. The patient was put on extracorporeal life support in the form of extracorporeal membrane oxygenator 12 h postoperatively due to low cardiac output and vasoplegic syndrome for 5 days. Later, his echocardiography showed significant left ventricular outflow tract obstruction in addition to severe pulmonary insufficiency which was confirmed by a diagnostic catheterization. Myectomy was performed to relieve the obstruction in the left ventricular outflow tract in addition to perventricular pulmonary melody valve replacement. Unfortunately, the patient developed significant mitral regurgitation,

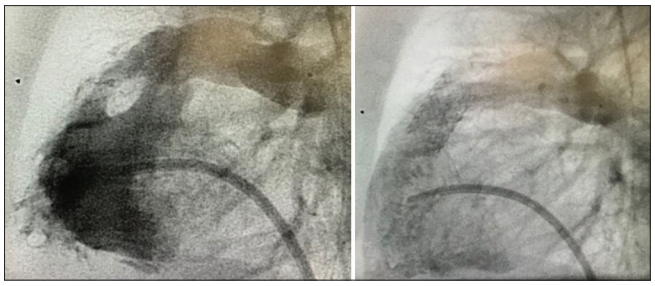

Figure 2: One of our patients with right ventricular outflow tract stenting

and we had to replace the mitral valve with bioprosthetic melody valve inserted surgically. The patient went home after a prolonged hospital stay and is doing fairly well one year after discharge.

Management of the RVOT obstruction follows the same strategies as in patients with isolated TOF. There is a detrimental effect of incompetent right AVV in the immediate postoperative period as these patients cannot tolerate both incompetent pulmonary valve and right AVV. Monocusp creation is meant to help the patient in the immediate postoperative period although all our series ended up by severe pulmonary regurgitation in the long term. On the long-term follow-up, it seems that severe pulmonary regurgitation is well tolerated by these patients. Excellent results have been demonstrated with both single- and double-patch techniques for the repair of AVSD.[4] It has been our policy to perform double-patch technique to repair the AVSD in all patients whether in isolated defect or if associated with TOF. We had only two patients with moderate to severe left AVV regurgitation after total repair. The first patient had a severe regurgitation preoperatively, and we had to replace the valve five days postoperatively with surgically implanted melody valve as mentioned earlier. The other patient had only moderate left AVV regurgitation postoperatively. This patient was managed medically and went home well. None of our cohorts had more than mild right AVV regurgitation.

Chylothorax is a frequent and serious complication associated with congenital heart surgery. It may be caused either by injury of the thoracic duct, increased pressure in the systemic veins exceeding that in the thoracic duct, or a central vein thrombosis.[11] While chylothorax may develop in virtually all types of intrathoracic procedures, several congenital heart operations have been shown to be prone to this condition, particularly bidirectional cavopulmonary shunt, Fontan type procedures, TOF, or AVSD repair.[12] The RV diastolic dysfunction after surgery for repair of TOF might increase the systemic venous pressure leading to the development of chylothorax.[12] The combination of the two lesions, i.e., TOF and AVSD repair increases the CPB time and cross-clamp time needed for repair. Biewer *et al.* found that the long duration of CPB and cross-clamp time are considered significant risk

factors for the development of chylothorax in addition to the low birth weight.[11] In our small cohort, the chylothorax occurred in 50% (n = 8) of patients, and although it might not be statistically significant, most of them were of the staged repair. None of our patients had a central venous thrombosis as a cause of chylothorax, and all of them were managed conservatively. No one needed surgical intervention.

The mean intensive care and hospital stay are in favor of the primary repair group, 3.5 and 16 days, respectively, compared to the staged repair cohort – 19.5 and 36 days. We have one case with long stay in the intensive care unit (ICU) due to complicated surgery and need for reintervention. This might explain the long ICU stay in the staged repair group. One should not forget that the primary repair group had more favorable RVOT anatomy while the staged group reflects the more severe and end of the spectrum of this complex disease.

Trisomy 21 was common in our cohort (94%). Brancaccio *et al.* showed 80% incidence of trisomy 21 patients with this disease which confirms the recent knowledge about the relationship between trisomy 21 and congenital heart defects.[13]

All literature which study these combined lesions have the limitation of small sample size due to the rarity of this disease. Meta-analysis might be advised.

CONCLUSION

CAVSD/TOF can be repaired with low mortality and morbidity. The use of RVOT stent has replaced the surgical (S-P) shunt with acceptable results in our center.

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Conflicts of interest

There are no conflicts of interest.

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