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ORIGINAL ARTICLE

Anatomic Repair of Congenitally Corrected Transposition of the Great Arteries: Single-Center Intermediate-Term Experience

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Abstract We present our experience for patients who have undergone an anatomic repair (AR) for congenitally corrected transposition of the great arteries (CCTGA) at the Children's Hospital of Wisconsin. A retrospective chart review of patients who underwent AR for CCTGA from 2001 to 2015 was performed. The cohort consisted of 15 patients (74% male). Median age of anatomic repair was 15 months (range 4.5–45.6 months). Four patients had a bidirectional Glenn (BDG) prior to AR. At the time of AR,—9 (60%) underwent Senning/Rastelli procedure, 4 (26.6%) had double switch operation, and 2 (13.3%) underwent only Senning with VSD closure. Median duration of follow-up was 5.5 years (0.05–14 years). Reoperations prior to discharge included BDG, revision of pulmonary venous baffle, closure of residual VSD, and pacemaker placement. Late reoperations included left ventricular outflow tract obstruction repair, conduit replacement, melody valve placement, and pacemaker implantation. At their most recent follow-up, no patient had heart failure symptoms and only 1 had severely diminished function that improved with cardiac resynchronization therapy. Moderate mitral regurgitation was noted in 15% (2/13), and severe in 7% (1/13). Moderate tricuspid regurgitation was noted in 15% (2/13). One patient, 7% (1/13), developed moderate aortic insufficiency. There was a 100% survival at the time of the most recent follow-up. Patients with CCTGA who have undergone AR have excellent functional status and mid-term survival but reinterventions are common. Longer term studies are needed to determine both the extent and spectrum of reinterventions as well as long term survival.

Keywords Transposition · Anatomic repair · Outcomes

Abbreviations

AR	Anatomic repair
BDG	Bidirectional glenn
CCTGA	Congenitally corrected transposition of the great arteries
MBTS	Modified Blalock–Taussig shunt
PA	Pulmonary artery
VSD	Ventricular septal defect

Introduction

Congenitally corrected transposition of the great arteries (CCTGA) is a rare cardiac malformation characterized by atrioventricular and ventriculoarterial discordance resulting from abnormal looping of the embryonic cardiac tube. The morphologic right ventricle receives inflow from the pulmonary veins and ejects to the aorta, and the morphologic left ventricle receives caval inflow and ejects to the pulmonary arteries [1]. The majority of these patients have associated cardiac defects, commonly ventricular septal defects (VSDs), pulmonary stenosis, pulmonary atresia, tricuspid regurgitation, and/or double outlet right ventricle. These patients can be present with heart failure or cyanosis in infancy or may have balanced circulations. Surgical interventions include either a physiological repair or an

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anatomic repair (AR). Physiologic repair involves correction of only the associated cardiac defects, leaving the morphologic right ventricle as the systemic ventricle. Patients with a physiological repair have a high rate of right ventricular dysfunction and tricuspid regurgitation over time compared to AR [2–4]. AR consists of redirection of blood flow at the atrial level with a Senning or Mustard procedure combined with either an arterial switch or a Rastelli procedure in order to place the morphologic left ventricle in the systemic position [5, 6]. Mid-term outcomes for an AR are encouraging, with a 70-80% survival rate at 10 years [2, 7–11].

Experience within the United States is limited, with only a few studies reporting more than 10 patients [6, 9, 12]. We present our intermediate-term experience for 15 patients who have undergone an AR for CCTGA at the Children's Hospital of Wisconsin, including a description of the patients' characteristics and postoperative course.

Patients and Methods

The Children's Hospital of Wisconsin Institutional Review Board approved the study and waived the need for parental or patient consent.

Patients

The cardiovascular surgical database at our institution (2001–2015) was used to identify 15 patients who had previously undergone an AR for CCTGA. A single surgeon performed all the surgeries in this cohort of patients. The surgeries performed included a double switch surgery (Senning-arterial switch), Senning-Rastelli, and only Senning with VSD closure. The individual surgical strategy was determined by a few key factors, including the associated abnormalities, particularly the nature of the VSD, and the presence or absence of pulmonary stenosis or atresia or right ventricular hypoplasia. None of the patients had preoperative arrhythmias or conduction abnormalities.

Double Switch Group (Senning-Arterial Switch)

Of the 15 patients, 4 (26.6%) underwent a double switch procedure. All 4 patients had levocardia with atrioventricular discordance and ventriculoarterial discordance ({S,L,L} segmental anatomy). A moderate-to-large perimembranous or outlet VSD was present in all 4 patients, without either pulmonary stenosis or pulmonary atresia.

Senning/Rastelli Group

Nine of the 15 patients (60%) underwent a Senning/Rastelli procedure. There were 3 patients with dextrocardia and 3 patients with heterotaxy syndrome. All of the patients had double outlet right ventricle with a moderate-to-large outlet type VSD. The majority of the patients (6/9, 67%) had {S,L,L} segmental anatomy and the remaining patients (3/9, 33%) had mirror image {I,D,D} segmental anatomy.

Senning with VSD closure

There were 2 of the 15 patients (13.3%) who underwent only a Senning procedure with VSD closure. Both of these patients had levocardia with atrioventricular discordance and ventriculoarterial concordance ({S,L,D} segmental anatomy) with a doubly committed VSD. There was no evidence of pulmonary stenosis in either of these two patients.

Data

A retrospective chart review of 15 patients who underwent AR for CCTGA from 2001 to 2015 was performed. Each patient's demographic data, surgical anatomy, type of anatomic repair (Senning-arterial switch, Senning-Rastelli, Senning with VSD closure) were identified. In addition, the need for and timing to reoperation/s, functional status, echocardiogram findings, and survival were evaluated at each patient's most recent follow-up.

Functional status was determined by their NYHA classification [13] in older children and Ross heart failure classification [14] in younger children. The echocardiogram features evaluated included ventricular systolic and diastolic function, valve function, and evidence of baffle obstruction. A single pediatric cardiologist assessed each patient's most recent echocardiogram.

Data analysis

To explore interrelationships and distributions, box and scatter plots and summary statistics such as the mean, median, range and correlation were used.

Results

The majority of patients in our cohort were male (11/15, 74%). At the time of AR, the median age was 16 months (range 4.5–45.6 months) and the mean weight was 9.8 kg (Table 1).

Table 1 Patient characteristics: this table describes patients demographics and the type of surgery performed at the time of AR

Patient characteristics	Patients (%)
Males	11/15 (73)
Heterotaxy syndrome	3/15 (20)
PA Band before AR	5/15 (33.3)
MBTS before AR	7/15 (46.6)
BDG before AR	4/15 (26.6)
Nature of AR	
Senning/Rastelli	9/15 (60)
Double Switch	4/15 (26.6)
Only Senning	2/15 (13.3)
VSD closure at AR	15/15 (100)
Glenn at AR	1/15 (6.6)

AR anatomic repair, *BDG* bidirectional Glenn, *MBTS* modified Blalock–Taussig shunt, *PA* pulmonary artery

Operative Details of AR

All 15 patients in our cohort underwent AR: 60% (9/15) underwent Senning/Rastelli, 26% (4/15) underwent double switch operation, and 13% (2/15) underwent Senning operation with VSD closure. A VSD was present in all of the patients (15/15) and was closed at the time of AR.

Senning-Arterial Switch Group (4/15 patients, 26%)

All of the patients in the Senning-arterial switch group underwent placement of a pulmonary artery (PA) band from 3 days to 3 months of life. Two patients with a large VSD and arch obstruction (coarctation of the aorta = 1 and interrupted arch = 1) underwent prophylactic placement of a PA band combined with arch repair in the first week of life. The other 2 patients received a PA band at 1 month and 3 months of age for relief of congestive heart failure. One patient had moderate mitral regurgitation going into AR. He was noted to have prolapse of his anterior and posterior mitral leaflets that was repaired with an Alfieri stitch at the time of the AR. Postoperatively, there was only mild mitral regurgitation.

Senning/Rastelli Group (9/15 patients, 60%)

One patient had aortic atresia and underwent a Norwood procedure in the first week of life, followed by bidirectional Glenn palliation (BDG). Of the remaining eight patients, seven had pulmonary atresia and underwent placement of a modified Blalock–Taussig–Thomas shunt (MBTS) in the neonatal period for ductal dependent pulmonary blood flow, followed by BDG (3/9 patients, 33%) or a second MBTS (3/9 patients, 33%) prior to AR. Earlier in our experience, we would target 1.5–2 years of age for Senning/Rastelli. The decision regarding a BDG versus a 2nd MBTS was based on the size of the morphologic RV. If the RV was mild to moderately hypoplastic, patients were staged to AR with a bidirectional Glenn shunt, whereas a second MBTS was chosen for those with a normal-sized RV. A second MBTS was common early in the experience although now we proceed with AR in infancy once the patient had outgrown the MBTS placed during the neonatal period. One patient who had a MBTS performed for neonatal palliation underwent BDG combined with AR due to concern for mild right ventricular hypoplasia. One patient with pulmonary stenosis did not require palliation and underwent primary AR at 4 years of age.

Senning with VSD Closure Group (2/15 patients, 13.3%))

One patient had an interrupted aortic arch with a large VSD and underwent arch reconstruction and PA band placement in first week of life. The second patient underwent primary AR at 6 months of age and that procedure also required VSD enlargement in order to create an unobstructed connection between the left ventricle and the aorta.

Reoperations

The majority of reoperations occurred prior to discharge following the initial AR (Table 2). Freedom from

Table 2 Reoperations: this table describes the early (prior to discharge) and late (after discharge) reoperations

Reoperation	Patients
Early (before discharge)	
Bidirectional Glenn	1/15 (6.6)
Revision of pulmonary venous baffle	2/15 (13.2)
Pacemaker placement	2/15 (13)
Closure of residual VSD	1/15 (6.6)
Late (after discharge)	
Conduit change	3/13 (23)
BDG take down	1/13 (7.6)
Melody valve	1/13 (7.6)
Repair of LVOTO	1/13 (7.6)
Pacemaker placement	1/13 (7.6)

BDG bidirectional Glenn, *LVOTO* left ventricular outflow tract obstruction, *VSD* ventricular septal defect

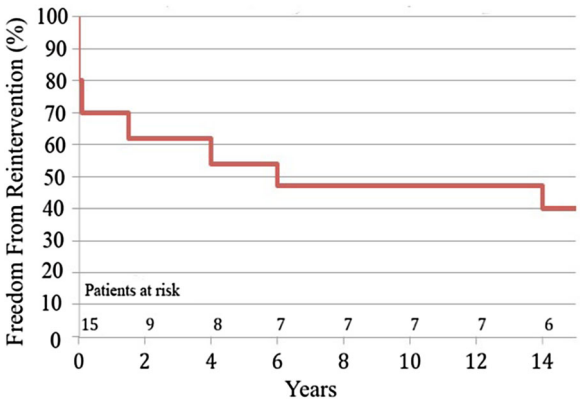

Fig. 1 Freedom from reintervention following anatomic repair

reoperation was 70% at 1 year and 47% at 10 years post AR (Fig. 1). There was no statistically significant association noted between any of the demographic variables or the nature of surgery prior to AR with nature of reoperation post AR.

Early Reoperations (prior to discharge after AR)

Average length of stay for all patients was 17 days (6–51 days).

Senning-arterial switch Group One patient required BDG in the immediate postoperative period due to superior vena cava baffle obstruction.

Senning/Rastelli Group One patient required pacemaker placement for postoperative complete heart block.

Senning with VSD closure Group Both patients underwent revision of the pulmonary venous baffle, one for baffle leak and the other for pulmonary venous baffle obstruction. One patient needed a pacemaker placement for postoperative complete heart block and closure of a residual apical VSD.

Late reoperations (after discharge from AR)

Two patients were lost to follow up with no access to their medical records following initial discharge from AR. Of the 13 remaining patients, the median duration of follow-up was 5.5 years (0.05–14 years).

Senning-arterial switch Group One patient developed severe left ventricular dysfunction 1 year after AR. This patient had a pacemaker placed for cardiac resynchronization therapy, which resulted in improved ventricular function.

Senning/Rastelli Group There were three patients who underwent RV to PA conduit changes from 1.5 to 6 years post AR. In addition to a RV to PA conduit, 1 of these 3 patients also required repair of left ventricular outflow tract obstruction and underwent elective takedown of BDG, as well as placement of a pacemaker for complete heart block at 4 years post AR. He subsequently required a Melody valve placement 14 years from the time of AR.

Senning with VSD closure Group There were no reoperations post discharge from AR in this group.

Functional Status

Survival was 100% at the time of the most recent follow-up. None of the patients have developed heart failure symptoms, with all patients NYHA class I or Ross heart failure class I at the time of their most recent follow-up. Ninety-two percent of patients (11/13) have normal biventricular systolic function and 1 patient has mildly diminished left ventricular systolic function. There was 1 patient with severely diminished left ventricular function, which has improved to mild to moderate dysfunction following pacemaker placement for cardiac resynchronization therapy. Mild mitral regurgitation was noted in 46% (6/13) patients, moderate in 15% (2/13—Senning only group), and severe in 7% (1/13—Senning/Rastelli group). The one patient with severe MR was the one with diminished LV function on his most recent study; otherwise MR was not associated with LV dysfunction and was structural in nature. Mild tricuspid regurgitation was noted in 38% (5/13) patients and moderate in 15% (2/13). No patients had severe tricuspid regurgitation. One patient, 7% (1/13) developed moderate aortic insufficiency. No patient had severe aortic insufficiency. Other than a RV to PA conduit, no interventions were directed at any of the valves post AR.

Discussion

Several studies have been performed to compare the 2 broad surgical approaches, physiological repair and anatomic repair (AR), to treat CCTGA. Although studies have demonstrated good early survival rates status post physiological repair, concerns remain regarding long-term functional outcomes [4, 7, 15]. Patients who undergo physiologic repair can develop systemic right ventricular dysfunction, especially in the setting of progressively worsening tricuspid regurgitation, which has been shown to be a risk factor for poor outcomes in patients with CCTGA [4, 12, 16–18].

In contrast, AR restores the left ventricle as the systemic ventricle, eliminating the systemic right ventricle, and would theoretically be a superior approach to this

condition. Ilbawi, et al. first reported AR as a surgical strategy for CCTGA in 1990, describing an atrial level switch combined with the Rastelli procedure in order to reposition the left ventricle in the systemic position [5]. Currently, AR typically includes either an atrial switch (Senning or Mustard) and arterial switch or a Senning and Rastelli procedure. [2, 5, 10, 19, 20]. Several studies from experienced centers worldwide have shown excellent results as far as long-term survival and quality of life [2, 8, 10, 21]. In addition, a meta-analysis by Alghamdi et al. showed that AR was associated with a significant improvement in the incidence of the in-hospital mortality compared to physiological repair [22]. However, due to its complexity, this surgery has not yet become a common procedure and physiological repair is still routinely performed at many institutions [4, 23]. In our institution, AR has been the preferred surgical approach for patients with CCTGA. Thus, given the limited published data in the United States regarding outcomes after AR, we present our surgical experience with AR for CCTGA.

At our institution, in order to make certain that the morphologic LV was prepared to do systemic work, AR was performed only if there was a large VSD. The type of AR repair to be performed was determined by the presence of pulmonary valvar stenosis and the nature and location of the VSD. Nine patients with severe pulmonary stenosis or pulmonary atresia underwent Senning/Rastelli surgery. There were two patients with a doubly committed VSD who underwent only a Senning with a left ventricular outflow tract baffle to the aorta and closure of the VSD. The remaining 4 patients underwent a double switch operation (Senning/arterial switch).

Palliative surgeries prior to AR were common, especially in patients who had varying degrees of right ventricular outflow tract obstruction. These patients underwent placement of a MBTS. Some of these patients underwent either a second MBTS or a BDG in anticipation of AR. A pulmonary artery band was used in patients with large VSDs to control pulmonary blood flow and protect the pulmonary vasculature. The use of pulmonary artery banding for left ventricular retraining has achieved mixed results [2, 3, 10, 24] and has been identified as a risk factor for postoperative mortality following the double switch [3]. No patient in this series underwent left ventricular retraining.

Studies have shown high rates of reoperation post AR. However, the reoperation rates are still favorable compared to patients who have undergone physiological repair [3]. In our study, freedom from reoperation was 70% at 1 year and 47% at 10 years, which is comparable to larger studies published [2, 3, 8]. The majority of the reoperations in our cohort occurred during the AR hospitalization. These included revision of the pulmonary venous baffle in the setting of leak or stenosis, BDG for superior vena cava baffle obstruction, or pacemaker placement for complete heart block. Both the patients with pulmonary baffle leak or stenosis had only undergone an isolated Senning with VSD closure. The reason for this is not entirely clear. However, obstruction and leakage of the pulmonary venous or systemic venous baffles requiring intervention is a recognized complication in the past [2, 3, 8, 9]. Importantly, the rate in our cohort was very low. Only 1 patient (6.6%) required reintervention for systemic venous baffle and 2 patients (13.3%) for pulmonary venous postoperatively. The higher rate of early as opposed to late reoperation reflects a learning curve of this complex operation. Based on our experience with early reoperations, we are more likely to augment the pulmonary venous pathway with in stiu pericardium. We have also learned that the SVC portion of the systemic venous pathway is more challenging in CCTGA, and are careful to resect all the atrial septum below the SVC right atrial junction and to reassess the SVC pathway during construction of the systemic venous baffle. Similar to other series of double switch operations, most of the late reinterventions in our series were directed at the RV to PA conduit [2, 3, 8].

Postoperative complete heart block needing pacemaker placement in the immediate postoperative period occurred in 2 patients (13%). One of those patients had undergone Senning/Rastelli and the other patient a Senning operation with VSD closure, which included VSD enlargement. This is different than past reports describing a higher incidence of complete heart block post double switch repair, where in addition to suture placement or VSD enlargement, traction on the crux of the heart was also noted to be a risk factor [3, 10]. One patient required pacemaker placement for complete heart block 4 years post Senning/Rastelli. It is important to remember patients with CCTGA remain at risk for compete atrioventricular block even without surgery. It has been estimated to affect approximately 2% of patients per year after diagnosis [25].

None of the patients in our cohort had severe tricuspid regurgitation or severe aortic insufficiency and only 1 patient had severe mitral regurgitation at their most recent follow-up. In this small cohort, these rates compare favorably with previous reports [2, 3]. None of our patients underwent interventions on the aortic or either atrioventricular valve post the initial AR.

We had 100% survival in our cohort at intermediate-term follow-up. Considering the anatomic complexity of the AR, high rate of reoperation/s, and suboptimal outcomes previously reported, some institutions are performing Fontan palliation on a subset of patients, with a lower rate of reoperation and a longer event free survival [11, 26]. While the intermediate results of single ventricle palliation

for CCTGA are favorable, these patients remain at risk for Fontan failure.

Right ventricular dysfunction has been noted in patients who have undergone physiological repair [12]. However, left ventricular dysfunction has been observed in patients post AR [27]. Only 1 patient in our cohort developed significant left ventricular dysfunction 1 year following AR consisting of double switch operation. This patient did have a pacemaker placed for cardiac resynchronization therapy, with considerably improvement in his function to mild to moderate dysfunction at his most recent follow-up. For patients with CCTGA and systemic ventricular dysfunction with a prolonged QRS or other evidence of dysynchrony, biventricular pacing should be strongly considered [28]. None of the patients underwent heart transplant. Importantly, all of the patients in our cohort had excellent functional status (NYHA or Ross class I) at most recent follow-up.

Limitations

This is a single-center retrospective study with a limited patient size and limited follow-up.

Conclusion

In conclusion, patients with CCTGA who have undergone anatomic repair have excellent intermediate-term functional status and survival, but reoperations are common. Further studies are needed to determine the long-term need for reoperations and survival.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

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