

CONGENITAL HEART DISEASE

ORIGINAL ARTICLE

Double Switch Procedure and Surgical Alternatives for the Treatment of Congenitally Corrected Transposition of the Great Arteries

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ABSTRACT ***Background:*** **We present our experience with the double switch operation in sixteen patients with congenitally corrected transposition of the great arteries.** ***Methods:*** **We enrolled 16 patients with congenitally corrected transposition of the great arteries operated by a single surgeon between 1995 and 2015. The mean age was 25 months (range 4 to 72 months) and the mean body weight was 8.9 kg (range 4.3 to 19 kg) at the time of operation.** ***Results:*** **We encountered seven patients with moderate to severe tricuspid regurgitation, five of which had Ebstein anomaly. We performed a combination of atrial and arterial switch procedures in 11 cases, one of which had a concomitant coarctation of the aorta that was repaired along with the double switch procedure. Atrial switch and the Rastelli procedures were performed in three cases with concomitant pulmonary stenosis. A combination of arterial switch, Hemi-Mustard procedure, and bidirectional cavopulmonary anastomosis was performed in two cases. During a mean follow-up period of 67 months (range three months to 18 years), we encountered one early postoperative mortality related to intracerebral bleeding. All but one of the patients are now in NYHA class I–II.** ***Conclusions:*** **Congenitally corrected transposition of the great arteries is a rare congenital cardiac anomaly in which the results of the anatomical repair with double switch operation appear to be superior to that achieved by a physiological repair.** doi: 10.1111/jocs.12728 *(J Card Surg 2016;31:231–236)*

Congenitally corrected transposition of the great arteries (cc-TGA) is a rare congenital cardiac malformation which is characterized by both atrioventricular and ventriculoarterial discordance. The surgical treatment options and timing of operation in cases with cc-TGA may be variable according to the associated congenital heart defects as well as the dominance of the left or right ventricle. Anatomical and physiological repair are the two main surgical options in cases with cc-TGA except for those with a hypoplastic ventricle in which Fontan circulation is mandatory.[1,2] The right ventricle becomes the systemic ventricle in the physiologic (i.e., classical or conventional) repair but the long-term outcome of this option is debatable.[3,4] Nevertheless, the double switch operation (DSO), which is also termed as the anatomical repair, results in the left ventricle serving as the systemic ventricle. The left ventricle may be trained with a prior pulmonary artery banding (PAB) followed by the DSO in selected cases.[5] Progressive impairment of function of the systemic right ventricle along with the regurgitation of the tricuspid valve leading to congestive heart failure is an inevitable result in cases treated with physiological repair.[6] On the other hand, in DSO, the left ventricle serves as the systemic ventricle, so the tricuspid valve and right ventricle are separated from the systemic circulation, and the long-term outcomes become more favorable than in the physiological repair.[7–13]

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In this report, we present our experience with the DSO in 16 patients with the diagnosis of cc-TGA in the last two decades. We also discuss long-term outcomes of the physiological and anatomical repair with a review of the literature.

MATERIALS AND METHODS

A retrospective analysis was performed in patients who underwent DSO with the diagnosis of cc-TGA following the approval of the local ethical committee and the institutional review board. Clinical presentations, preoperative echocardiographic findings, perioperative course, and follow-up records were evaluated.

Sixteen patients (13 male, 3 female) with cc-TGA were operated on by a single surgeon between 1995 and 2015. Mean age was 25 months (range: 4 months to 72 months) and mean body weight was 8.9 kg (range 4.3 to 19 kg). All of the patients were evaluated by transthoracic echocardiography and thirteen of the patients underwent cardiac catheterization before the operation. The demographic and operative profiles of the patients are presented in Table 1.

Surgical technique

Cardiopulmonary bypass with moderate to low flow hypothermia was implemented following aortic and bicaval cannulation. Following cross clamping of the aorta, cold antegrade blood cardioplegia was administered and intermittent doses were given every 15 minutes. Hemofiltration was implemented during the warming period on cardiopulmonary bypass. An autologous pericardial patch was treated in glutaraldehyde solution (with a concentration of 0.5%) for three minutes. We performed three types of surgical techniques:

1. ***Atrial and Arterial Switch Procedure (group 1) (n = 11):*** Atrial switch (Senning procedure) and arterial switch procedures were performed in this group where the atrial switch was completed prior to the arterial switch operation. In cases where the atrial septal defect was large, the interior flap was constituted with a polytetrafluoroethylene patch. The outer flap was enlarged with autologous pericardium. The pulmonary venous baffle was reconstructed with in situ pericardium, which was reported as Barrat-Boyes modification and was performed in two cases.[13] In patients who had a concomitant ventricular septal defect, the defect was closed via the transatrial route followed by the arterial switch operation.
2. ***Atrial Switch and Rastelli Procedure (group 2) (n = 3):*** The ventricular septal defect was closed after the Senning procedure was completed. Three types of extracardiac conduits (16 mm Dacron; 21 mm Freestyle™ Bioprosthesis, Medtronic, Minneapolis, MN, USA and 18 mm Contegra™ Pulmonary Valved Conduit, Medtronic, Minneapolis, MN, USA) were used in three cases in order to provide right ventricle to pulmonary artery continuity.
3. ***Arterial Switch and Hemi-Mustard Procedures, Bidirectional Cavopulmonary Anastomosis (group 3) (n = 2):*** In this group, the arterial switch operation was performed prior to the Hemi-Mustard operation and the procedure was completed with the anastomosis of the superior vena cava to the right pulmonary artery.

We did not perform a Lecompte maneuver in any of the cases. The mean cardiopulmonary bypass and aortic cross-clamp times were 199 ± 32 and 153 ± 17 minutes, respectively. All of the patients were hemodynamically stable with the sternum closed when they left the operating room except for one patient who was placed on a systemic ventricular assist device for eight days.

RESULTS

All of the patients had situs solitus. A concomitant ventricular septal defect (VSD) was present in all cases. We encountered 13 patients with levocardia, two patients with dextrocardia and another one with mesocardia. Five of the patients had coronary artery anomalies, three of which had a single coronary root. We encountered two patients with Wolff-Parkinson-White syndrome and one patient with complete atrioventricular block.

We performed a combined atrial and arterial switch procedure in 11 cases (group 1). A prior pulmonary artery banding (PAB) was performed in three cases (pt. no. 6, 8, and 16) in this group in order to train the left ventricle. The DSO was performed 2 weeks, 5 months, and 18 months after the PAB procedure. Ebstein anomaly was present in five cases and two other cases had moderate to severe tricuspid valve regurgitation in this group. In all cases with tricuspid valve incompetence, the regurgitation was observed to regress after the DSO. We repaired a concomitant aortic coarctation in the same session with DSO, performing the resection of the coarctation with an end to end anastomosis. Patient no. 16 was admitted with the symptoms of a low cardiac output state associated with severe right atrioventricular valve regurgitation. He was put on a temporary (left atrium to ascending aorta) ventricular assist device after the DSO procedure for eight days from which he was successfully weaned.

We operated on three patients with cc-TGA and associated pulmonary stenosis in which we performed an atrial switch and a Rastelli procedure (group 2). One patient in this group with pulmonary stenosis was palliated by a prior systemic to pulmonary artery shunt. The pulmonary arterial anastomosis was partially shifted onto the left pulmonary artery in order to prevent aortic compression.

Two patients with the diagnosis of cc-TGA, pulmonary stenosis, and right ventricle dysfunction, one of which had Ebstein anomaly, were treated with arterial switch and hemi-Mustard procedures with a concomitant bidirectional cavopulmonary anastomosis (group 3). In one case with a significant compression of the pulmonary artery, a 19 mm graft (Vascutek™,

TABLE 1
Demographic, Operative, and Long-Term Outcomes

Patient	Age (months)	Body Weight (kg)	Associated Cardiac Pathology	Surgical Procedure Group 1, 2, 3*	Long-Term Outcomes
1	4	4.3	VSD, PDA	Group 1, PDA closure	Early postoperative mortality
2	5	4.9	VSD	Group 1	Mild to moderate AR, NYHA I
3	6	5.2	VSD, TR	Group 1	NYHA I
4	72	14	VSD (multiple)	Group 1	Mumps infection related LV dysfunction, NYHA II
5	49	9.6	VSD, Ebs, PS, TR, AV block	Group 3, PS repair, PM implantation	NYHA II
6	6	5	VSD, Ebs, TR, s/p PAB	Group 1, pulmonary debanding	Late Cx artery occlusion, NYHA I
7	18	10	VSD, Ebs, TR, coA	Group 1, coA repair	Mild to moderate AR, NYHA I
8	36	12	VSD, Ebs, TR, s/p PAB	Group 1, pulmonary debanding	NYHA I
9	48	16	VSD, PS	Group 2	Reoperation for early RV-PA conduit thrombosis, NYHA I
10	4	4.1	VSD	Group 3	Reoperation for pulmonary venous baffle stenosis, RF ablation, mild to moderate AR, NYHA I
11	60	18	VSD, PS	Group 2	NYHA III, scheduled for dual PM implementation
12	50	14	VSD, PS, s/p SPS	Group 2, division of the shunt	RV-PA conduit replacement, NYHA I
13	24	9.2	VSD, TR	Group 1	NYHA I
14	7	6.2	VSD	Group 1	NYHA I
15	5	5.1	VSD	Group 1	NYHA I
16	6	5	VSD, Ebs, TR, s/p PAB	Group 1, pulmonary debanding	Postoperative temporary mechanical circulatory support, mild to moderate AR, NYHA I

*Group 1: Atrial and arterial switch operation; group 2: atrial switch and Rastelli procedure; group 3: arterial switch and hemi-Mustard procedures, bidirectional cavopulmonary anastomosis.
VSD, ventricular septal defect; PDA, patent ductus arteriosus; TR, tricuspid regurgitation; Ebs, Ebstein anomaly; AV, atrioventricular; PS, pulmonary stenosis; PAB, pulmonary artery banding; CoA, coarctation of the aorta; SPS, systemic to pulmonary shunt; PM, pace-maker; RV, right ventricle; PA, pulmonary artery; AR, aortic regurgitation; LV, left ventricle; Cx, circumflex coronary artery; RF, radiofrequency ablation.

Terumo Corp., Tokyo, Japan) was interposed to the ascending aorta. The ventricular septal defects were closed through the mitral valve in group 1 or through the ventriculotomy in cases in which a Rastelli procedure was performed.

Early postoperative surgical reintervention was necessary in three cases. One patient was reoperated on three days after the DSO due to pulmonary venous baffle obstruction and the outer flap was enlarged with a polytetrafluoroethylene patch (pt. no. 2). Thromboembolectomy was performed in one patient in group 2 (pt. no. 9) who had thrombosis of the right ventricle to pulmonary valved conduit (Contegra^{TM}) one month after the operation. We implanted a permanent epicardial pacemaker in one patient with complete atrioventricular block (6.6%) who was in sinus rhythm until the fifteenth postoperative day (pt. no. 14). We encountered one patient with temporary atrioventricular block in group 1 who spontaneously returned to sinus rhythm ten days after the operation. A permanent pacemaker was placed during the double switch operation in patient no. 5 who had preoperative complete atrioventricular block.

We encountered four cases with long-term complications in the follow-up period, two of which underwent reoperation. One patient was reoperated eight years after an atrial switch and Rastelli procedure in order to revise a Dacron conduit stenosis with interposition of a homograft between the right ventricle and the main pulmonary artery. In another patient with pulmonary venous baffle stenosis and Wolff-Parkinson-White syndrome, the pulmonary venous pathway was enlarged and the radiofrequency ablation was performed eight years after the operation. This patient had a single supraventricular tachycardia episode four years after the reintervention. In patient no. 4, the ejection fraction decreased to 45–50% after an episode of mumps infection several years after the atrial and arterial switch operations. This was thought to be due to myocarditis secondary to the mumps. Cardiac catheterization revealed a normal coronary circulation in this patient. This patient now has NYHA class II symptoms. We encountered a late coronary arterial obstruction in which the circumflex artery was filled via retrograde flow and the circumflex coronary artery was demonstrated to have a separate ostium during surgery. Although this patient has mild mitral and aortic regurgitation, the left ventricle ejection fraction and tread mill test are within normal limits. In the follow-up period, eight patients are free of neoaortic regurgitation.

Four patients have mild and another four have mild to moderate neoaortic regurgitation. We did not encounter any progressive aortic valve regurgitation. The overall late left ventricular ejection fraction is maintained at 55% to 65% which is based on regular transthoracic echocardiographic evaluations.

Mortality was encountered in one case (6.6%), which was related to postoperative cerebral hemorrhage (pt. no. 1). Although this patient had a successful cardiac repair confirmed by transthoracic echocardiography in the intensive care unit, he never regained consciousness after the operation. The computed tomography revealed extensive cerebral hemorrhage and the patient died on the eleventh postoperative day. We did not encounter any late mortality in a mean follow-up of 67 months (range 3 months to 18 years).

All patients are now in NYHA Class I–II, except for one young adult (pt. no. 11). Atrial switch and Rastelli procedures were performed in this patient and a permanent epicardial pacemaker was implanted due to atrioventricular block. He has been followed for 17 years and the recent transthoracic echocardiography revealed biventricular dysfunction. We scheduled a dual pacemaker implantation for this patient.

DISCUSSION

Congenitally corrected transposition of the great arteries is a rare cardiac malformation which has a unique pathology as well as variable clinical presentation even in the seventh decade of life.[14] The natural history and clinical presentation of the cases with cc-TGA are closely related to the coexistence of a ventricular septal defect, which is reported to be the most commonly associated congenital heart defect in these patients.[1] In our series, all of the patients had concomitant ventricular septal defects, one of which was multiple (pt. no. 4). Pulmonary outflow obstruction is associated in half of the cases and more than 90% of this subset of patients have Ebstein anomaly of the tricuspid valve.[15,16] We encountered five (31.2%) patients with Ebstein anomaly of the tricuspid valve. Two of our patients (13.3%) had pulmonary outflow stenosis and five patients (33.3%) had a coronary anomaly.

Although the long-term outcome of the surgical treatment with conventional repair in patients with cc-TGA is questionable, anatomical repair provides a better long-term outcome with regard to ventricular and systemic valvular functions, since the left ventricle becomes the systemic ventricle whereas the right ventricle becomes the pulmonary ventricle as in normal physiology. Although the DSO is a more challenging technique, it is associated with lower mortality and mortality when the long-term results are considered.[17] Karl and associates reported a similar initial operative risk for the anatomical and conventional repair; however, the DSO was superior in long-term follow-up.[18] On the other hand, Shin'oka and colleagues reported that long-term results are similar for the conventional repair and the DSO, but they suggested performing the double switch operation in cases with tricuspid regurgitation.[8]

The major limitation of the conventional repair is right ventricle dysfunction and tricuspid regurgitation which eventually leads to congestive heart failure. This has resulted in a 17% ten-year mortality, 50% right ventricle dysfunction, and 71% moderate to severe tricuspid valve incompetence according to the data reported by Sano and colleagues.[19] Tricuspid valve function is noticed to be significantly improved in the follow-up period after the DSO.[12] In our patient population with the anatomical repair, the long-term results are more satisfactory when compared to outcomes with the conventional repair.

In neonates presenting with cc-TGA and associated ventricular septal defect, PAB is an important treatment strategy. This surgical option is associated with improved tricuspid valve function in terms of regurgitation, which is probably related to the modification of the septal geometry following the banding procedure.[20] Moreover, PAB provides the training of the morphological left ventricle in these cases. The left ventricle mass/volume ratio is a reliable marker for the timing of the DSO after PAB.[21] In our opinion, a left ventricle mass/volume ratio more than 1.5 is a dependable criteria for the exact timing of DSO after PAB and we used this management strategy in three of our patients. We observed significant improvement in tricuspid valve competence after PAB in these patients, one of whom was operated on with ventricular failure (pt. no. 16). We operated on three patients (pt no: 6, 8, and 16) whose ages were ranging between 6 and 36 months in order to perform a PAB followed by the anatomical repair for cc-TGA. Although we did not encounter any older patients with cc-TGA, we had an experience with an adolescent with simple TGA, who had undergone PAB in another cardiac surgery center at the age of 5. In our evaluation, the morphological left ventricle was not ready for the arterial switch procedure, therefore we reoperated on him in order to tighten the pulmonary band, and then proceeded with the arterial switch procedure at the age of 10. He is 21 years old now and his left ventricle function is normal without any evidence of neoaortic regurgitation.

Myers and colleagues presented the midterm results of PAB in their cc-TGA patient population. The age at PAB procedure ranged between 15 days to 23 years and the period for left ventricular training after PAB was between 2 weeks and 11 years.[22] On the other hand, Poirier and associates emphasized that the response of the morphological left ventricle to training with a PAB procedure is less favorable in their patients over the age of 12.[23] Nevertheless, they state that the inconsistent response of the left ventricle to training is a challenging problem especially in patients over 20 years of age.

Pulmonary and systemic baffle obstruction are well-known complications of the atrial switch procedure.[24] We encountered an early and a late pulmonary venous baffle obstruction in two patients in our population (13.3%), three days and eight years after the double switch operation, respectively. Atrioventricular block is

an important clinical problem in patients with cc-TGA, since the conduction system is morphologically abnormal in cases with situs solitus.[25] Moreover, the rhythm disturbances that are encountered after the atrial switch procedure as well as the technical challenges during the closure of the ventricular septal defect increases the risk of atrioventricular block in the postoperative period. Sharma and colleagues pointed out the importance of atrial arrhythmias encountered in the follow-up period of the DSO, and reported that six of their patients (8.8%) had atrial rhythm disturbances after the DSO and two of them died due to intractable atrial arrhythmias.[26]

Our experience suggests that the anatomical repair with the DSO is superior than the conventional technique in patients with cc-TGA Moreover, in our opinion, cc-TGA patients with tricuspid valve regurgitation and Ebstein anomaly should not be conventionally repaired, since the right ventricle and tricuspid valve which become the systemic ventricle and valve in classical repair fail and will eventually end up with congestive heart failure on long-term follow-up. Pulmonary artery banding is still a reasonable option in two subgroups of the patients with cc-TGA: newborns presenting with an associated ventricular septal defect and patients whose left ventricle should be trained before the DSO. Moreover, PAB is effective for decreasing the degree of tricuspid regurgitation before the DSO.[12] In cases with unbalanced ventricles, univentricular palliation with the Fontan circulation is the appropriate treatment option.

Although cc-TGA is a rare cardiac malformation, its treatment mandates meticulous surgical technique as well as a strict follow-up schedule. We believe that the anatomical repair is the first choice in surgical treatment and results in more favorable long-term outcomes.

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