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ORIGINAL ARTICLE

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A better approach for left ventricular training in transposition of the great arteries and intact interventricular septum: Bidirectional cavopulmonary anastomosis and pulmonary artery banding

Mehmet Salih Bilal MD[1] | Arda Özyüksel MD[1,2] | Mustafa Kemal Avşar MD[1] | Şener Demiroluk MD[3] | Osman Küçükosmanoğlu MD[4] | Yalım Yalçın MD[5]

[1]Department of Cardiovascular Surgery, Medicana International Hospital, Istanbul, Turkey

[2]Department of Cardiovascular Surgery, Biruni University, Istanbul, Turkey

[3]Department of Anesthesiology, Medicana International Hospital, Istanbul, Turkey

[4]Department of Pediatric Cardiology, Medicana International Hospital, Istanbul, Turkey

[5]Department of Pediatric Cardiology, Florence Nightingale Hospital, Istanbul, Turkey

Correspondence
Arda Özyüksel, Department of Cardiovascular Surgery, Medicana International Hospital, Beylikdüzü Caddesi, No:3, Beylikdüzü, Istanbul, Turkey.
Email: ozyukselarda@yahoo.com

Abstract

Objective: Management of the patients with transposition of the great arteries and intact ventricular septum may be challenging beyond the newborn period. Herein, we would like to present our alternative strategy for training the left ventricle in these patients.

Methods: Six patients with transposition of the great arteries and intact ventricular septum were evaluated in our clinic. Two of them were palliated with Glenn procedure and pulmonary banding as a definitive treatment strategy at other centers. Four patients were operated on and a bidirectional cavopulmonary anastomosis in combination with pulmonary artery banding was performed (stage-1: palliation and ventricular training) in our center. In four out of these six patients, arterial switch operation was performed with takedown and direct re-anastomosis of the superior vena cava to right atrium after an interstage period of 21-30 months (stage-2: anatomical repair).

Results: Any mortality was not encountered. The left ventricular mass indices increased from 18-32 to 44-74 g/m^2 in patients undergoing the anatomical repair. All of the patients were uneventfully discharged following the second stage. The mean follow-up period was 20 months (9-32 months) following stage 2. All of the patients are doing well with trivial neoaortic regurgitation and normal biventricular function.

Conclusions: Bidirectional cavopulmonary anastomosis with pulmonary artery banding may be a promising left ventricle training approach in ventriculoarterial discordance when compared to the traditional pulmonary artery banding with concomitant systemic-to-pulmonary artery shunt procedures which still carry a significant interstage morbidity and mortality.

KEYWORDS

arterial switch operation, bidirectional Glenn procedure, intact ventricular septum, modified Blalock–Taussig shunt, transposition of the great arteries

1 | INTRODUCTION

D-looped transposition of the great arteries (TGA) accounts for 5%-7% of congenital heart defects.[1] Surgical outcomes of the arterial switch operation (ASO) within first 4 weeks of life is encouraging; however, the clinical situation becomes more troublesome beyond this period.[2] The atrial switch procedures were the principal form of management for late presentations of patients with TGA for decades.[3] In fact, the atrial switch procedures, namely the Senning and Mustard operations convert TGA to a situation similar to congenitally corrected transposition of the great arteries (cc-TGA) where the right ventricle and the tricuspid valve serve for the systemic ventricle and atrioventricular valve, respectively. However, recognized long-term complications include systemic right ventricular dysfunction, brady and tachyarrhythmias, baffle obstructions, and even sudden death.[3-5]

In the absence of a source of volume and/or pressure load on the left ventricle (LV), most commonly a patent ductus arteriozus (PDA) or ventricular septal defect (VSD), the ability of the LV to sustain a systemic function begins to decrease gradually.[6] Beyond the neonatal period, preparing or "retraining" the LV for ASO in cases with TGA is mandatory when the interventricular septum is intact (TGA-IVS). In traditional approach, pulmonary artery banding (PAB) and/or systemic-to-pulmonary artery shunt procedures are preferred, however, these procedures have significant morbidity and mortality.[7] The mortality of this first step of a two-stage ASO may be as high as 15% due to complications such as low cardiac output, hypoxemia necessitating a secondary intervention for shunt resizing, aortic regurgitation, decrease in pulmonary band gradient in the early postoperative period.[8,9] The intensive care unit stay may be complicated with unsuccessful extubation and prolonged necessity of inotropic support. Moreover, this stormy postoperative course may lead to increased morbidity and mortality which may even overcome the risks of ASO.[10]

Herein, we would like to present an alternative left ventricular training procedure for such cases in which we perform bidirectional cavopulmonary anastomosis (Glenn procedure) along with PAB. The subsequent successful ASO procedures were performed with takedown of the cavopulmonary anastomosis and debanding of the pulmonary artery in four out of six cases.

2 | PATIENTS AND METHODS

Six patients were evaluated with the diagnosis of TGA-IVS beyond the infancy period in our clinic. The ages of the patients at the presentation were varying between 5 months and 3 years. Two of them were operated on at foreign centers in order to palliate the patients aiming to increase systemic saturation levels. Four patients were operated and a bidirectional cavopulmonary anastomosis along with PAB were performed in our center. Overall, these six patients were defined as stage-1 palliation group. Five of these patients had secundum atrial septal defects as the sources of mixing between

TABLE 1 Summary of the patients who underwent anatomical repair following stage-1 palliation

Patient no.	Age/BW at stage-1	Age/BW at stage-2	Interstage period	Technique of Glenn takedown	LV mass index at stage-1 (g/m^2)	LV mass index at stage-2 (g/m^2)	RV/LV ratio at stage-2	Inotropic support after stage-2	Result/follow-up period
1	5 mo/4 kg	2 yrs/12 kg	21 mo	Direct SVC-RA anastomosis	23	74	0.79	None	Well/32 mo
2	8 mo/5 kg	3 yrs/13 kg	27 mo	Direct SVC-RA anastomosis	25	44	0.91	Adrenaline and milrinone	Well/28 mo
3	9 mo/5 kg	3.5 yrs/16 kg	29 mo	Direct SVC-RA anastomosis	18	52	0.77	None	Well/14 mo
4	13 mo/7 kg	4 yrs/15 kg	30 mo	Direct SVC-RA anastomosis	32	59	0.94	Milrinone and dobutamine	Well/9 mo

Abbreviations: BW, body weight; CPB, cardiopulmonary bypass; LV, left ventricle; RA, right atrium; RV, right ventricle; SVC, superior vena cava.

oxygenated and deoxygenated blood and one patient had a history of balloon atrial septostomy at the newborn period.

Targets of stage-1 palliation were determined as a loose PAB for left ventricular preparation and bidirectional cavopulmonary anastomosis for providing adequate systemic oxygenation. Patent ductus arteriosus were ligated with silk sutures in two patients at the first stage. All the patients were operated via midline sternotomy, and standard hemodynamical parameters were achieved at the end of Glenn anastomosis and PAB. Glenn anastomosis was performed with veno-venous shunting between the superior vena cava (SVC) and right atrium (RA), without implementing cardiopulmonary bypass. An appropriately sized venous cannula was inserted to SVC, preferably near the junction with the innominate vein. Another venous cannula was inserted to RA. When the SVC was clamped during the anastomosis, blood flow to the RA was easily preserved by this technique. On the other hand, the Glenn procedure was performed first, followed by PAB. This provided a safe and more accurate way for tightening the pulmonary band when adequate systemic oxygen saturation and ventricular pressure ratios were concerned. The target systemic oxygen saturation in blood gas analysis was 80%-90% at a FiO_2 of 50% and a tidal ventilation volume of 10-12 ml/kg. The targeted pulmonary band gradients and mean main pulmonary artery pressures were 30-40 mm Hg and 15 mm Hg, respectively. All of the patients were uneventfully discharged after stage-1 palliation. Two of the recent stage-1 patients are being followed up for training their LVs. Out of six stage-1 patients, four of them were reoperated for stage-2 anatomical repair. The interstage period was 21, 27, 29, and 30 months for these patients, respectively. All of the patients underwent cardiac catheterization before stage-2 anatomical repair. The left ventricular mass indices were calculated to be 74, 44, 52, and 59 gr/m^2, respectively.

Stage-2 anatomical repair integrated pulmonary artery debanding, takedown of the previously constructed SVC to right pulmonary artery and re-anastomosis of the SVC to RA and ASO. All of the SVC to RA anastomosis were directly performed following adequate mobilization of the vessel without any need for graft interposition. The pulmonary reconstruction following the debanding procedure was performed in synchronization with neopulmonary reconstruction component of the ASO. A bovine pericardial patch was used for this purpose. All of the coronary arteries were in usual pattern and they were transferred to the neoaorta with separate buttons. Le Compte maneuver was performed in all cases. The atrial septal defects were closed with bovine pericardial patches. The patients who underwent anatomical repair following stage-1 palliation are summarized in Table 1.

3 | RESULTS

Postoperative follow-up following the second-stage repair was uneventful except Patient 2. This patient, who had the lowest LV mass index at stage-2 repair in our patient population (44 g/m^2) was weaned from CPB without inotropic support and the intraoperative transthoracic echocardiography revealed good biventricular function. However, in the sixth postoperative hour, adrenaline and milrinone infusions were started due to the decreased left ventricular ejection fraction (35%-40%) and mild to moderate neoaortic valve regurgitation with findings of low cardiac output. The patient's ventricular function gradually recovered after 72-96 hours. He was extubated after 7 days and transthoracic echocardiography revealed LVEF: 55% with mild aortic regurgitation at the time of discharge. Patient 4 received prophylactic milrinone and dobutamine at the termination of CPB, which were gradually ceased in the follow-up.

All of the patients were discharged uneventfully following the second-stage definitive repair. Postoperative transthoracic echocardiography evaluations were scheduled at regular intervals. The mean follow-up was 17 months (5-28 months) and all of the patients are doing well in this period.

4 | DISCUSSION

The history of surgery for TGA has paralleled the history of cardiac surgery. Moreover, attempts to palliate cyanotic newborns with TGA even began with Blalock–Hanlon septectomy which was performed in 1948, before the era of open-heart surgery.[11] In the following decade, the atrial switch was defined in order to physiologically correct the transposition. Afterward, the untoward effects of atrial switch were sought to be addressed by ASO which aimed to anatomically correct TGA.[12] Today, the ASO is currently the procedure of choice for TGA, especially when performed in the first month of life.[13] However, in many parts of the world, a significant number of the newborn patients with TGA cannot be diagnosed at the early period and alternative strategies are deemed mandatory for treatment of these infants.[14]

In cases where the LV is exposed to systemic pressures from birth such as in cases with unrestrictive VSD, pulmonary stenosis, congestive heart failure or pulmonary hypertension, it sustains the systemic pressures after the arterial switch procedure, which is a similar fact following double switch procedures in patients with cc-TGA, as well.[15] The LV can be trained to support the systemic circulation, which is traditionally performed by applying a pulmonary artery band to increase its afterload and stimulate its hypertrophy along with a systemic-to-pulmonary artery shunt in order to maintain an acceptable systemic saturation level.[13,16] However, the postoperative course may be stormy in these patients, especially in terms of intensive care unit stay, delayed extubation, need for inotropic support. Although the modified Blalock–Taussig shunt (mBTS) is the most commonly performed technique for providing systemic to pulmonary artery blood flow, it continues to be associated with high operative death, reintervention and interstage mortality risks. Petrucci and colleagues reported the risks for mBTS with regard to the outcomes of the Society of Thoracic Surgeons database which showed an operative mortality of 7.2%.[17] On the other hand, a report published by Alsoufi presents a hospital mortality of 14.9% for mBTS and only 5% mortality in 2 years after Glenn procedure.[18]

The same report states that unplanned reoperations after mBTS are as common as 13.8% and this situation may increase the operative mortality from 14% to 21%.[18] In their histopathological analysis of 155 shunts, Wells and colleagues found that at the time of takedown, the mean value for maximal narrowing of the shunt lumen was 34 ± 22% and 21% of the patients had stenosis more than 50%. The shunt size less than 4.0 mm had significantly increased risk for stenosis, which may be a concern in almost all patients at the infancy period.[19] Increased shunt size to weight ratio is another significant risk factor for overcirculation and subsequent coronary steal.[18] Therefore, it's clear that an associated mBTS in such an LV training strategy may significantly decrease the overall success of the ASO.

In such a query where the untoward effects of mBTS are evident, the creation of a bidirectional cavopulmonary anastomosis (BDG) may have several advantages. As the major component of first-stage palliation in patients with TGA-IVS, the pulmonary banding serves as a way of increased afterload for the LV. Sun and colleagues reported a statistically significant mean decrease of LV ejection fraction, global strain and lateral wall strain in patients undergoing PAB for LV training in patients with ventriculoarterial discordance (TGA) with or without atrioventricular discordance (cc-TGA).[20] Moreover, these measures of LV function showed more but a statistically insignificant decrease following the arterial or double switch procedures for TGA and cc-TGA, respectively.[20] Therefore, the decremental effects of an mBTS may become more evident in terms of coronary steal. Such a disadvantage is not a point of question for BDG. There are two important immediate effects of BDG physiology.[21] First, the pulmonary recirculation is eliminated with concurrent volume unloading of the heart. Second, the BDG is a more efficient model for oxygenation when compared to mBTS since there is systemic rather than admixed venous blood flow to the pulmonary circulation.[21] This aids to provide a more stable hemodynamic status by improving effective systemic output. The volume unloading of the heart does not seem to have a significant adverse effect on training the LV, since pressure overload exposes more contractile and elastic reserves of the ventricle than volume overload in animal models.[22] These advantages of BDG over mBTS may improve the outcomes both at the interstage period and after ASO procedure in patients with TGA. On the other hand, the younger age of our patient population may have positive effect on the results as expected in these circumstances. Any technical challenge was not encountered when the takedown of the Glenn anastomosis and re-anastomosis of the SVC to RA is considered in stage-2. In fact, we did not re-anastomose SVC to the original site at the right atrium (the cavo-atrial junction) where the right atrial stump was over sewn during the Glenn procedure. Instead, we performed a right atriotomy superior and caudal to the cavoatrial junction, and resected the pectineal muscles in the atrial wall. Afterward, re-anastomosis of the SVC was uneventfully performed (following adequate mobilization of the vessel) to this site, which is similar to the anastomosis line in Warden Procedure. We also did not encounter any problem related to the diameter of the SVC in our patient population.

Several topics with regard to the optimal surgical management strategy for TGA-IVS are concerned since the two-stage repair (PAB and training of LV followed by ASO) was proposed by Sir Magdi Yacoub in 1977.[23] Jonas and colleagues presented the concept of rapid two-stage anatomical repair, in which the interstage period was significantly short (5-7 days).[24] More creative solutions such as PDA recanalization/stenting.[10] administration of levosimendan and continuous positive airway pressure,[2] adaptable PAB systems,[25] different PAB strategies applied in animal models (ie, permanent, progressive or intermittent)[26] along with the availability of mechanical circulatory support[27] extended the limits of ASO in patients with TGA-IVS beyond the neonatal period even up to 9 months of age.[28]

A recent clinical guideline highlighted the challenging issues in treatment strategies for TGA-IVS[13] Currently, the optimal timing for an ASO in newborns with TGA-IVS is accepted to be within the first days of life up to 3 weeks of life.[29] However, for infants up to 2 months of age, a primary ASO may be considered with mechanical circulatory back-up; whereas, a two-stage approach ASO will be preferred for the patients beyond 2 months. The noninvasive criteria for the need for LV training include LV mass index <35 g/m^2, age>3 weeks, and "banana shaped" LV on 2D-echocardiography. Three types of LV training techniques are outlined in this report; (1) PAB with mBTS, (2) PAB with induced patency of the arterial duct, obtained either by prostaglandin infusion or stenting, and (3) induced patency of the arterial duct alone, obtained either by prostaglandin infusion or stenting.[13] A postbanding LV/RV ratio of 0.65-0.70 is recommended. Proposed criteria for a safe second-stage ASO include (1) LV/RV ratio > 0.85, (2) LV end-diastolic volume > 90% of normal, (3) LV ejection fraction > 50%, (4) posterior wall thickness > 4 mm, and (5) LV wall stress < 120 × 10^3 dynes/cm^2.[13,30] Lacour-Gayet F and colleagues perform the ASO when the LV mass index had reached 50 g/m^2.[7]

Our stage-1 palliation strategy with PAB and BDG enabled a more silent postoperative course. We preferred a looser pulmonary banding and LV/RV pressure adjustment strategy in order to provide a smooth training period for the LV with an acceptable extension over a period of time. We adjusted the pulmonary band in order to achieve a 30-40 mm Hg gradient along with a LV/RV pressure ratio of 0.5. We aimed to provide a systemic oxygen saturation above 85% with the combination of PAB and BDG. The pulmonary artery band was loosely constructed at the initial surgery with a Gore-Tex or silk tape, which could be retightened afterward in order to increase pressure load of the LV. However, we did not retighten the pulmonary band in any of our patients. Our aim was to provide a loose rather than tight band with 3 presumptions:

1. The postoperative course following stage-1 procedure (Glenn + PA banding) was more stable in terms of hemodynamics and respiratory parameters, earlier extubation and more rare necessity for inotropic support. The patients were even ready to be transferred to the ward in the first postoperative day, with satisfactory systemic oxygenation, oral intake, urinary output, and cardiac performance.

2. Prolonging the training period following the initial palliation. In fact, these patients were admitted to our center from Middle East and African countries. Although follow-up and timing of second-stage repair are meticulously scheduled for these patients, delays in readmission may be encountered. In such patients, any unexpected delay for stage-2 definitive repair does not have noteworthy effect in quality of life, morbidity, or mortality.
3. The possibility of a relatively tighter effect of the banding material as the child grows up.

The postoperative course was uneventful for all of the patients. The interstage period was between 21 and 30 months for these patients who were traveling from abroad. The follow-up during the interstage period included both subjective and objective parameters. The parents were questioned about the general condition and growth of the child as well as any symptoms if present. The objective parameters included effort capacity, ventricular function, valvular competence, pulmonary band gradients, interventricular septal geometry, left ventricular mass index, and any other echocardiographic findings if present. At the second admission to our hospital, all of the patients were evaluated with cardiac catheterization and any early development of arteriovenous fistula or pulmonary artery distortion were not encountered. The parents were questioned about the quality of life and physical performance of the patients at the interstage period and all were doing well with satisfactory mental and physical development and exercise tolerance when they began walking. The LV mass indices and LV/RV pressure ratios were adequate for ASO at their second admission, which were all completed uneventfully.

TGA with intact ventricular septum is a challenging clinical scenario in advanced patient age in terms of effective but uneventful staged repair is concerned. Unfortunately, late referral of the patients with TGA is still an important problem in underdeveloped countries. Our observation and experience in classical training with a shunting procedure (important morbidity and mortality overwhelming the arterial switch procedure itself) encouraged us to develop an alternative strategy, especially in older patients. In patients who are admitted with TGA-IVS between 2 weeks and 3 months of age, we adopt the classical rapid two-stage repair (pulmonary banding and systemic to pulmonary artery shunt followed by anatomical repair). On the other hand, we prefer this alternative approach with pulmonary banding and Glenn procedure in patients older 3 months of age as stage-1 palliation.

We believe that the hibernated-like left ventricular myocardium may effectively and safely be trained for anatomical repair with pulmonary banding and bidirectional anastomosis. This approach in our limited patient series may provide a better and acceptable systemic oxygen level, improved ventricular blood supply, less stormy postoperative course, and a more uneventful interstage training period when compared to the traditional approaches.

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interest with the content of this article.

AUTHOR CONTRIBUTIONS

MSB: Concept/design, data analysis, critical revision of the article.
AÖ: Concept/design, drafting the article, approval of the article.
MKA: drafting the article, data analysis, data collection.
ŞD: drafting the article, data analysis.
OK: data analysis, critical revision of the article.
YY: Concept/design, data analysis, critical revision of the article.

ORCID

Arda Özyüksel https://orcid.org/0000-0001-7478-6235

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