Late Correction of Transposition of the Great Arteries

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ABSTRACT

Introduction: Surgical correction of transposition of the great arteries (TGA) is usually performed in the first week of life. There is no consensus on surgical intervention for patients with a late diagnosis. Our study was designed to evaluate the early results of patients who underwent late-term correction based on surgical techniques.

Patients and Methods: Our study included patients older than a month, who were operated on due to TGA between 2014 and 2019. Data on the postoperative hospital mortality and morbidity of these patients were examined.

Results: The study enrolled 11 patients. Arterial switch operation (ASO) was performed in 36.3% (n= 4) of the patients, while an atrial switch operation was performed in 63.7%. After left ventricular (LV) training, ASO was performed in 25% (n= 1) of the patients. Extracorporeal membrane oxygenation (ECMO) support requirement was present in 36.4% (n= 4) of the patients due to postoperative low cardiac output. The mortality rate was 27.3% (n= 3). Of these deaths, 33.3% occurred among patients who had undergone Senning operations, while 66.7% occurred among those who underwent ASO (n= 1). While 66.7% (n= 2) of the mortality was due to low cardiac output, 33.4% was due to sepsis (n= 1).

Conclusion: There is still no absolute consensus on the timing of surgical intervention in cases of TGA. ASO can be performed in cases with an increased need for ECMO and acceptable mortality among patients over one month of age. For patients with LV regression, two-stage ASO and atrial switch operations are alternative options.

Key Words: Transposition of the great arteries; Arterial Switch operation; Senning operation; left ventricular training

Büyük Arter Transpozisyonunda Geç Dönem Korreksiyon

ÖZET

Giriş: Büyük arter transpozisyonunun (TGA) cerrahi tamiri genellikle yaşamın ilk haftasında gerçekleştirilir. Geç tanı almış hastalarda, cerrahi konusunda fikir birliği yoktur. Çalışmamızda geç dönem korreksiyon yapılan hastaların cerrahi tekniğe göre erken dönem sonuçlarını değerlendirmeyi amaçladık.

Hastalar ve Yöntem: Çalışmamıza 2014-2019 yılları arasında TGA nedeniyle opere edilen ve yaşı bir aydan büyük olan hastalar dahil edildi. Hastaların postoperative hastane mortaliteleri ve morbiditeleri incelendi.

Bulgular: Çalışmaya 11 hasta dahil edildi. Hastaların %36.3'üne (n= 4) arterial switch operasyonu (ASO), %63.7'sine atrial switch operasyonu yapılmıştır. Sol ventrikül (LV) training sonrası ASO %25'inde (n= 1) gerçekleştirildi. Postoperative düşük kardiyak output nedeniyle hastaların %36.4'üne (n= 4) ekstrakorporal membran oksijenasyonu (ECMO) desteği gerekti. Mortalite %27.3 oranında gözlenmiştir (n= 3). Bunların %33.3'si Senning operasyonu (n= 1), %66.7'si arterial switch yapılan hastalardı. Mortalitenin %66.7'si (n= 2) düşük kardiyak debi, %33.4'ü sepsis nedeniyle gerçekleşti (n= 1).

Sonuç: Büyük arter transpoziyonunda cerrahi zamanlaması için halen kesin bir yargı yoktur. Arterial switch operasyonu yaşı bir ayın üzerinde olan hastalarda artmış ECMO ihtiyacı ve kabul edilebilir mortaliteyle gerçekleştirilebilir. LV regrese olan hastalarda iki aşamalı ASO ve atriyal switch operasyonu diğer seçeneklerdir.

Anahtar Kelimeler: Büyük arter transpozisyonu; Arteryel Switch operasyonu; Senning operasyonu; sol ventrikül training

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INTRODUCTION

Late diagnosis of congenital heart disease is a common problem in developing countries[1]. As a consequence of this, increased age was found to be associated with high morbidity and mortality in complete anatomical correction. TGA is a typical example of this, and late diagnosis is still common among these patients[1,2].

TGA is observed with an incidence rate of 0.2 per 1000 live births worldwide[3], and it is one of the cyanotic heart diseases that require emergency surgical interventions. Surgical outcomes are usually associated with the timing of the surgery[4]. The mortality rate among untreated patients with TGA in the first year of life is 90%[5]. The arterial switch operation (ASO) performed within the first two to three weeks is the ideal surgical approach for today's cases of TGA with an intact ventricular septum[6]. In most studies, the safe age limit for primary ASO is considered to be 21 days, and two months is considered to be a possible age limit with an increased need for extracorporeal membrane oxygenation (ECMO). There are limited studies on patients undergoing surgery after two months, and none of those previous studies were statistically significant[7]. ASO and atrial switch operations after left ventricular (LV) training with pulmonary banding are other alternative options for patients beyond the neonatal period[2,8]. This study was undertaken with the aim of sharing our surgical experiences with TGA patients who underwent operations after one month of age.

PATIENTS and METHODS

Our study enrolled 11 patients older than a month, who were operated on due to TGA between 2014 and 2019. This retrospective cross-sectional study was approved by the local ethics committee (Date: 12.01.2021, Decision no: 2021/01/420). Group A included patients who underwent one-stage or two-stage ASO, while Group B included those who underwent atrial switch operations.

Early morbidity and mortality rates were evaluated in relation to the ages of patients at the time of operations and the procedures performed. Prolonged cross-clamp time, age and saturation at the time of operation, prolonged ventilation in the postoperative period, delayed sternal closure, need for peritoneal dialysis or ECMO, length of intensive care and hospital stay, and early mortality rates were compared.

Statistical Analysis

Variables were analyzed using IBM SPSS Statistics 276.0 (IBM Corp., Armonk, NY, USA). Conformity of the data to normal distribution was evaluated with the Kolmogorov-Smirnov test. In the comparison of the two independent groups based on quantitative data, the Mann-Whitney U test was used together with the Monte Carlo results. In the comparison of categorical variables with each other, the Fisher exact test and Monte Carlo simulation technique were applied. Quantitative variables were expressed as mean (± standard deviation) and median (minimum-maximum) in tables, while categorical variables were shown as n (%). The variables were analyzed at the 95% confidence level and values of $p< 0.05$ were considered significant.

RESULTS

The demographic characteristics of the patients were reviewed in detail (Table 1). The mean age was 40.36 ± 52.01. Of the patients, 45.5% had a history of previous interventions. ASO was performed in 36.4% of the patients, Senning operation in 54.5%, and Mustard operation in 9.1%. The rate of application of two-stage ASO was 27%. In 54.5% (n= 6) of cases, simultaneous ventricular septal defect closure was performed. ECMO was required in 36.4% (n= 4) of the patients. Of these, 66.3% (n= 2) were patients in whom ASO was performed, while 33.7% (n= 1) had undergone a Senning operation. The mortality rate was observed to be 36.3% (n= 4). Of these deaths, 50% occurred among patients who had undergone a Senning operation, while 50% occurred among those who underwent ASO (n= 2). While 75% (n= 3) of deaths occurred due to low cardiac output, 25% occurred due to sepsis (n= 1).

Distributions of patients based on their diagnoses, operative findings, and postoperative complications were evaluated separately and are presented in Table 2. The effect of ASO or atrial switch operation on postoperative morbidity was also compared (Table 3); no difference was found between the two groups. Previous history of operation was also observed to not affect postoperative mortality (Table 4).

DISCUSSION

Atrial switch operations were first introduced by Senning and Mustard[9,10] and began to be performed routinely in the 1970s[11]. When the ASO was subsequently introduced, it was quickly adopted despite the good survival rate after the atrial switch[12]. It is thought that when the morphological left ventricle is a systemic ventricle, the systemic ventricular and atrioventricular valve dysfunction seen in the late period after atrial switch operations can be prevented[13,14]. Atrial switch operations in cases of TGA with ventricular septal defect (VSD) give poor early results. For this reason, ASO is preferred after the neonatal period[15,16].

Surgical repair is usually performed with low mortality in the first week of life, between the third and seventh days. The preferred approach is ASO. Septal defects are repaired simultaneously with other possible cardiac and extracardiac defects[17]. When performed after one week, ASO is a risk factor for myocardial dysfunction due to decreased adaptation of the left ventricle to high pressure.

Table 1. Demographic distribution of patients

	n	%
Gender		
Female	3	27.3%
Male	8	72.7%
Diagnosis		
TGA-VSD	7	63.6%
TGA	3	27.3%
TGA-VSD-PS	1	9.1%
Previous Intervantion/Surgery		
Balloon septostomy	2	18.2%
Pulmonary banding	2	18.2%
Modified Blalock-Taussig shunt	1	9.1%
None	6	54.5%
Surgery		
ASO	4	36.4%
Mustard operation	1	9.1%
Senning operation	6	54.5%
Additional procedure		
VSD closure	6	54.5%
Rastelli operation	1	9.1%
None	4	36.4%
Postoperative complication		
Atrial baffle revision	2	18.2%
Low cardiac output (LCO)	2	18.2%
Pneumothorax	1	9.1%
LCO-Subdural bleeding	1	9.1%
Sepsis	1	9.1%
AV block-prolonged ventilation	1	9.1%
Tracheoesophageal fistula	1	9.1%
None	2	18.2%
Two-Stage ASO		
(-)	8	72.7%
(+)	3	27.3%
ECMO		
(-)	7	63.6%
(+)	4	36.4%
Mortality		
(-)	8	72.7%
(+)	3	27.3%

Table 1. Demographic distribution of patients (continue)

	n	%
Coronary anomaly		
(-)	9	81.8%
(+)	2	18.2%
Peritoneal dialysis		
(-)	9	81.8%
(+)	2	18.2%
	Mean (SD)	**Median (Min/Max)**
Age	40.36 (52.01)	24 (1/156)
Weight	10.95 (8.78)	8 (3.1/30)
ICU stay	27.82 (37.57)	8 (1/100)
Hospital stay	35.55 (42.93)	15 (1/133)

ASO: Arterial switch operation, AV block: Atrioventricular block, TGA: Transposition of the great artery, VSD: Ventricular septal defect, PS: Pulmonary stenosis, LCO: Low cardiac output, ECMO: Extracorporeal membrane oxygenation, ICU: Intensive care unit.

In a study conducted by Anderson et al., it was suggested that the length of hospital stay and mortality rate increased with surgical interventions after the fifth day of life[4].

Regarding the timing of surgery, retrospective studies have focused on operations performed in patients with late diagnoses[18,19] or operations performed a few hours after delivery[20,21]. The research conducted by Anderson et al. at two medical centers in North America found that the ideal timing for ASO was the third day of life[4]. In a study conducted by Cain et al., similar results were obtained for patients operated on after the seventh day (between the eight and 14th days)[22]. In another study, it was shown that good results were achieved even for those patients who underwent ASO after one-two months and for those who had left ventricular regression and moderate pulmonary hypertension[23].

The surgical options for patients with TGA-IVS and patients older than two-three weeks are primary ASO, two-stage ASO, and atrial switch procedures following pulmonary banding[24]. Recently published guidelines indicate that primary ASO may be performed with class IB indications for up to three weeks and with class IIA indications for up to two months[25].

In a study published in 2020, which enrolled patients operated on after the eighth week, Daoud et al. found that the mortality rate was 0% and early ECMO was needed in 45% of the cases. Postoperative systemic ventricular dysfunction, on the other hand, was detected in 90.9% of patients.

Among the findings obtained in another previous study, no change was observed in long-term results among patients who underwent ASO after three weeks. Improved LV function was stable after surgery. Banana-like appearance or depressed LV function was observed in the left ventricle in all patients operated on after three weeks, six weeks, and two months. However, the frequency of ECMO increased with age. LV mass and posterior wall thickness were shown to be important in determining the preoperative status of the left ventricle, but it was concluded that these could be ignored in determining the surgical strategy for patients who underwent surgery after 21 days[26]. In addition, it has been suggested that LV mass is not a predictor of postoperative ventricular function and good clinical results in such cases[27].

For patients with TGA-VSD or Taussig-Bing anomaly, ASO can also be performed at an early stage with the intent of avoiding early and rapidly developing pulmonary vascular disease. However, in this group of patients, the period is not considered late until six months after birth[28,29].

In our study, we performed ASO and simultaneous VSD closure in three patients aged one month, four months, and four months. Two of these patients needed ECMO due to postoperative low cardiac output, and one of them died. In our clinic, the prevailing opinion is that if the surgical operation is planned to be performed within the first six months, it is necessary to perform ASO and simultaneous closure of the intracardiac defect. It is thought that this can be performed in appropriate patients who have relatively higher morbidity and mortality risks.

Most authors recommend a two-stage ASO after one month. This technique aims at LV adaptation for high pressure[30,31].

Table 2. Detailed characteristics of patients

No	Age (Month)	Gender	Weight (kg)	Diagnosis	Previous Operation	Surgery	Additional Procedure	Two-Stage ASO	ICU Stay (Day)	Postoperative Complication	Hospital Stay (Day)	ECMO	Mortality	Coronary Anomaly	Peritoneal Dialysis
1	156	Male	30	TGA-VSD	Pulmonary banding	Mustard	VSD closure	(+)	26	Pneumothorax	36	(-)	(-)	(-)	(-)
2	24	Male	8	TGA-VSD	Pulmonary banding	ASO	VSD closure	(+)	100	LCO- Subdural bleeding	133	(+)	(-)	(-)	(+)
3	24	Male	10	TGA-VSD	None	Senning	VSD closure	(-)	94	Sepsis	94	(-)	(+)	(-)	(-)
4	12	Male	6	TGA	Baloon septostomy	Senning	None	(-)	3	None	15	(-)	(-)	(+)	(-)
5	3	Female	4	TGA	None	Senning	None	(-)	4	Tracheoesophageal fistula	7	(-)	(-)	(+)	(-)
6	4	Female	4	TGA-VSD	Baloon septostomy	ASO	VSD closure	(-)	9	LCO	9	(+)	(-)	(-)	(+)
7	4	Male	4.3	TGA-VSD	None	ASO	VSD closure	(-)	1	LCO	1	(+)	(+)	(-)	(-)
8	1	Female	3.1	TGA-VSD	None	ASO	VSD closure	(-)	54	AV Block-Prolonged ventilation	60	(-)	(-)	(-)	(-)
9	36	Male	11	TGA-VSD-PS	Modified BT-Shunt	Senning	Rastelli Operation	(+)	8	Atrial Baffle Revision	8	(+)	(+)	(-)	(-)
10	60	Male	18	TGA-VSD	None	Senning	None	(-)	2	Atrial Baffle Revision	7	(-)	(-)	(-)	(-)
11	120	Male	22	TGA	None	Senning	None	(-)	5	None	21	(-)	(-)	(-)	(-)

ASO: Arterial Switch operation, AV block: Atrioventricular block, TGA: Transposition of great artery, VSD: Ventricular septal defect, PS: Pulmonary stenosis, LCO: Low cardiac output, ECMO: Extracorporeal membrane oxygenation, ICU: Intensive care unit.

Table 3. Comparison of the patients according to the surgical technique

	Surgery		
	ASO	Senning op	
	(n= 4)	(n= 6)	p
Gender (Male), n (%)	2 (50.0)	5 (83.3)	0.500[f]
Age, median (min/max)	4 (1/24)	30 (3/120)	0.132[u]
Weight, median (min/max)	4.15 (3.1/8)	10.5 (4/22)	0.067[u]
ICU stay, median (min/max)	31.5 (1/100)	4.5 (2/94)	0.476[u]
Hospital stay, median (min/max)	34.5 (1/133)	11.5 (7/94)	0.762[u]
Postoperative complication, n (%)	4 (100)	4 (66.7)	0.467[f]
Two-stage ASO, n (%)	1 (25)	1 (16.7)	0.999[f]
ECMO, n (%)	3 (75.0)	1 (16.7)	0.190[f]
Mortality, n (%)	1 (25.0)	2 (33.3)	0.999[f]
Coronary anomaly, n (%)	0 (0.0)	2 (33.3)	0.467[f]
Peritoneal dialysis, n (%)	2 (50.0)	0 (0.0)	0.133[f]

[u] Man-Whitney U test (monte carlo), [f] Fisher exact test (Monte Carlo).
ASO: Arterial switch operation, ECMO: Extracorporeal membrane oxygenation, ICU: Intensive care unit.

Table 4. Comparison of reoperation and postoperative complications and mortality

	Previous Surgery		
	(-)	(+)	p
Postoperative complication			
(-)	1 (16.7)	1 (20.0)	0.999
(+)	5 (83.3)	4 (80.0)	
Mortality			
(-)	4 (66.7)	4 (80.0)	0.999
(+)	2 (33.3)	1 (20.0)	

Fisher exact test (Monte Carlo).

LV training was first described by Yacoub et al. in 1977[32]. Jonas reported the fast two-stage switch technique in 1989[24]. Lacour-Gayet et al. described the indications as age >1 month, a banana-shaped left ventricle, LV mass of <35 g/m^2, interventricular septum bulging right to left, and low LV pressure. Corno et al., Wernovsky et al., and Jonas et al. found significant increases in LV mass index, RV/LV wall thickness ratio, LV/RV pressure ratio, LV mass and volume, and mass/volume ratio after retraining[24,30,33]. Two-stage ASO should be performed when the LV mass index is 50 g/m^2, the LV pressure has increased, and the interventricular septum has been shown to be flattened[23].

It has been suggested that a rapid two-stage ASO must be performed following the demonstration of LV hypertrophy after pulmonary banding. ASO was performed a week later. In the early period, low operative mortality and normal LV functions were observed[24]. During pulmonary banding, the LV/RV ratio should be kept between 0.33 and 0.6-0.7[28]. When the mortality rates of patients who underwent LV training after three months were compared, the mortality rates were found to be 85.7% at six months, 66.7% at one year, and 66.7% at five years. Death after LV training was observed in patients over three months of age[23].

In our study, only one patient underwent two-stage ASO, and he was one year old. This patient was treated with ECMO due to postoperative DCO. In addition, due to subdural hematoma, there was a need for extended durations of mechanical ventilation and intensive care stay. The patient was discharged

in good health. Performing LV training after six months of age was associated with postoperative complications and mortality. Fast two-stage surgery can be more effective for some patients.

For patients with a late presentation, the Senning operation is an important alternative. The Senning protocol should be preferred for patients with satisfactory RV and tricuspid valve functions who have problems during the preoperative period[34]. The Senning/Mustard procedure may also be indicated in the presence of low birth weight, late diagnosis, anomalies that prevent implantation of the coronary artery, and dysplastic pulmonary valve. ASO conversion is controversial because some patients show RV dysfunction that is not associated with coronary artery disease in the late stage. However, this indication can be considered in cases of patients who are at a high risk of LV dysfunction after the pulmonary band. Recent studies suggest that ASO conversion should be considered when there is late RV failure and when medical treatment is ineffective[35]. Although atrial correction provides good early results, a long-term increase in RV pressure is associated with a long-term risk of morbidity and mortality. There was also an increased risk of atrial arrhythmia due to a large number of incisions in the atria[36]. In their study, Talwar et al. investigated the mid-term results of atrial switch operations in patients over five years of age. It has been claimed that some modifications can help reduce the incidence of baffle obstruction and arrhythmia[37]. For a successful Senning operation, Schumacker's modification is recommended. In the classical Senning procedure, the autologous atrial septum is used with the intent of separating the pulmonary venous return from the mitral valve. However, the absence of the interatrial septum is critical in older patients. Therefore, it is recommended to use a Dacron patch in the reconstruction of the systemic venous baffle with the Mustard technique. It is possible to prevent long-term baffle stenosis and pulmonary venous obstruction with Schumacker's in situ pericardial technique. There are concerns about the possibility that this technique may cause sinus node dysfunction and ventricular dysfunction in the long term[38]. Another strategy is the gradual preparation of the left ventricle and additional application of a loose band to the pulmonary artery during Senning operations to prevent right ventricular dysfunction. In short, atrial switch procedures with their good early and mid-term results are still valuable techniques for the treatment of patients with late presentation[37].

In our study, atrial switch operations were performed for 63.7% of the patients. In our clinic, the Senning operation is preferred in cases where the patient is over one year of age, preoperative comorbidity is present, severe LV dysfunction is observed through echocardiography, and concomitant coronary anomalies and severe desaturation are present. In our study, ECMO was needed for a patient treated with simultaneous Senning and Rastelli operations, who developed a postoperative atrial baffle leak. Two occurrences of postoperative atrial leakage were observed, and the use of Dacron patches during baffle treatment is thought to be useful in minimizing this. Mustard operation was recommended for a patient with a history of permanent pacemaker implantation since the leads strongly adhered to the right atrium.

CONCLUSION

An ASO is the first choice despite the increased morbidity and mortality rates after one month among patients diagnosed with TGA. Fast LV training can be an option for eligible patients. A Senning operation is a safe choice for those with serious preoperative comorbidities or those older than one year.

Ethics Committee Approval: The study protocol was approved by the Kartal Koşuyolu High Specialization Training and Research Hospital Ethics Committee. The study was conducted in accordance with the principles of the Declaration of Helsinki (Decision no: 2021/1/420 Date: 12.01.2021).

Informed Consent: This is retrospective study, we could not obtain written informed consent from the participants.

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