

A 15-Year Experience with an Old but Still Challenging Operation: the Systemic-Pulmonary Artery Shunt

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Abstract

Objective- The true incidence of congenital cardiovascular malformations is difficult to determine accurately, partly because of difficulties in definition. About 0.8 percent of live births are complicated by a cardiovascular malformation. Hypoxia and cyanosis, the common complications of all cyanotic disease, may be life-threatening in severe forms. Today, the trend is towards the total surgical correction of these anomalies in early life. As the accomplishment of this strategy in various parts of the world is not possible, palliative procedures like systemic-pulmonary shunt have retained their importance.

Methods- Data were collected from the files of 180 patients, for whom systemic-pulmonary shunt was performed by a single surgical group at our center between March 1992 and May 2006. Our aim was to determine the outcome of shunt operation in terms of success rate, morbidity, and mortality.

Results- The median age and weight of the patients was 24 months and 10.5 kilograms, respectively. There was a spectrum of underlying cyanotic heart diseases. The main operation was the modified Blalock-Taussig shunt (90%). The mean value of oxygen saturation was 62% pre-operatively, which rose to 85% after surgery. We found a 77.9% success rate, 6.7% mortality rate, and 8.7% morbidity rate.

Conclusion- There was no significant correlation between the predictive factors and success of operation. Lower age and weight of the patient, small size of the pulmonary artery, and urgency of operation predicted the operative mortality (*Iranian Heart Journal 2008; 9 (3):10 -17*).

Key words: cyanosis ■ systemic-pulmonary shunt ■ central shunt ■ tetralogy of Fallot

Despite progress in medical sciences in recent decades, heart diseases are among the most prevalent causes of mortality and disability in human beings. About 0.8% of live births are complicated by a cardiovascular malformation, of which 20% are cyanotic.¹ Common to all cyanotic heart diseases are anatomic and hemodynamic features, by which intracardiac shunts and diminished pulmonary blood flow lead to hypoxemia and cyanosis.

heart disease might have led to the innovation of shunt surgery.^{2,3}

Congenital heart surgery includes the palliative treatment and surgical complete repair of cardiac malformations in newborns, children, and adolescents.

Palliative surgery allows early or long-term survival, depending on the primary malformation and the condition of the patient. Establishment of a systemic-to-pulmonary shunt (the modified Blalock-Taussig shunt)

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allows, in general, recovery from severe cyanosis and leads to the growth of the hypoplastic pulmonary vascular tree in newborns with severe pulmonary stenosis.⁴

Nowadays, despite the efforts all around the world for an early correction of cardiac anomalies, shunt surgery is still commonly performed and has its role in the treatment of these patients.^{5,6} Systemic-pulmonary shunt remains a major strategy for the palliation of cyanotic lesions in neonates despite the associated morbidity and mortality⁷. A better understanding of the anatomy and physiology of congenital heart disease has improved pre- and post-operative care; and deep hypothermia, circulatory arrest, and miniaturization of the equipment, among other factors, have contributed to the greatly increased safety of open-heart surgery in neonates and infants. Consequently, a trend towards an early correction has developed, which prompts the question: "In which congenital heart anomalies presenting early in life should primary repair be preferred to initial palliation followed by late repair?" It is imperative to weigh the advantages and disadvantages of a two-stage "palliative + corrective" procedure against primary correction.⁵

On the other hand, in certain cases, due to the presence of allocated cardiac or other congenital lesions, a palliative shunt operation may be the first line of therapy until further stabilization and more assessment of underlying malformations can be achieved.⁸

Confronting such diseases as hypoplastic left heart syndrome (HLHS) or pulmonary atresia in the neonatal period, we must perform a procedure which includes systemic-pulmonary shunt as well. Therefore, understanding the key points and pitfalls of the shunt operation is extremely important for the practicing surgeon.

In light of the above-mentioned facts, we decided to perform a cross-sectional study and assess the results of the systemic-pulmonary shunt surgery in our center. The

purpose of this study was to identify the outcome of the shunt operation.

Methods

One hundred and eighty patients underwent systemic-pulmonary shunt between March 1992 and May 2006 at our institution. The mean age was 44.5 months (median, 24 months; range, 1 to 120 months), and the mean weight was 13.23 kg (median, 10 kg; range, 3 to 36 kg). There were 54.4% male and 45.6% female patients.

The underlying lesions were comprised of severe tetralogy of Fallot, including tetralogy with pulmonary atresia (n=67), complete transposition with pulmonary stenosis (n=37), pulmonary atresia (n=22), single ventricle with pulmonary stenosis (n=23), tricuspid atresia (n=17), and double outlet right ventricle with severe pulmonary stenosis (n=14).

All the patients with significant cyanosis who underwent systemic - pulmonary shunt were enrolled in the study. There were four exclusion criteria: 1-neonates with complete transposition who underwent the modified Blalock-Taussig shunt and Pulmonary artery banding as part of a rapid two-stage arterial switch, 2-all neonates with hypoplastic left heart syndrome treated by the Norwood procedure, 3-babies with pulmonary atresia who underwent a simultaneous valvotomy and/or right ventricular out-flow tract augmentation, and 4-patients with associated intracardiac procedures in addition to a systemic-pulmonary arterial shunt.

Technique of operation

The operative technique did not differ from that generally used in the creation of a modified Blalock-Taussig shunt (MBTS), classic Blalock-Taussig shunt (CBTS), and central shunt.

Classic Blalock-Taussig shunt was done according to the traditional technique of this procedure in the early years of our study only.

In modified Blalock-Taussig shunt (MBTS) via posterolateral thoracotomy, the subclavian artery and left or right pulmonary artery are dissected. After heparinization with a dose of 1-1.5 mg/kg, a longitudinal incision is made in the subclavian artery near its origin, and a cobra-head anastomosis is performed with the obliquely cut polytetrafluoroethylene (PTFE) tube. For the distal anastomosis, the left or right main pulmonary artery is incised longitudinally or transversely. By a proper measurement of the tube graft length, anastomosis is performed with a single running suture of 6-0 polypropylene.⁹⁻¹¹

In the central shunt, the sternum is split vertically and part of the thymus is removed, and the pericardium is opened in its cephalic portion. A careful evaluation of the anatomy of the main pulmonary artery and its branches, especially their diameters, determines the site of shunt outflow. The innominate artery or ascending aorta serves as the shunt in-flow site. After a sharp dissection of the corresponding arteries, systemic-pulmonary shunt is established using a PTFE tube in the heparinized patient.

After the operation, all the surviving patients were mechanically ventilated for a variable length of time, maintaining peripheral oxygen saturations at more than 85%.

On arrival in the intensive care unit (ICU), all the patients underwent echocardiographic examinations to visualize the shunt and establish early baseline patency for comparison with subsequent examinations.

Statistical analysis

The data were expressed as mean or median (SD=standard deviation, expressed in parentheses) for interval and frequency (%) for the categorical variables. The independent sample *t*-test was used to compare the means of the interval variables between the groups. The Mann-Whitney U-test was used to compare the mean of the interval variables without normal distribution or ordinal data between the two groups. For the categorical

data, comparisons between the groups were carried out using the Pearson Chi-square test. P-values less than 0.05 were considered statistically significant.

A multivariate analysis was performed to determine the adjusted associations between the major outcomes of the study (success, mortality, and morbidity of operation) and other predictors, including: age, sex, weight, underlying diseases, pulmonary artery diameter, surgical incision type, urgency of operation, and oxygen saturation before surgery. The categorical variables with more than two categories were converted to dummy variables. A logistic regression model was used for the success of operation, and the Poisson regression models were fitted for the morbidity and mortality outcomes.

STATA 8 SE (STATA Corp., Texas, USA) was used for statistical analysis.

Results

Baseline data

As mentioned previously, 180 patients, 83 female (45.9%), with a median (SD) age of 24 (46) months (range, 1 - 196 months) and median (SD) weight of 10.5 (10) kilograms (range, 3 - 75 kilograms) were enrolled in the study. The mean values (SD) of the partial pressure of oxygen and oxygen saturation before surgery were 38 (8.0) mmHg and 62 (14.7) percent, respectively. The diameter of the pulmonary artery was very small (z -value < -2) in 32 (17.7%), medium ($-2 < z$ -value < 0) in 107 (59.1%), and normal (z -value ≥ 0) in 42 (23.2%) cases. The main operative technique was modified Blalock-Taussig shunt (MBTS), performed in 162 (89.5%) patients. The classic Blalock-Taussig shunt was performed in 3 (1.7%) and central shunt in 16 (8.8%) other patients. The surgical incision was located on the right side in 116 (64.1%), left side in 48 (26.5%), and mid-sternotomy in 17 (9.4%) patients. The tube graft used in the patients was of different sizes, the majority used (84.3%) being 5 or 6 mm in diameter; and 15.4% of the grafts were

4 mm or less. In the majority of the cases (92.3%), the operation was performed on an elective basis. The median values (SD) for ICU stay, hospital stay, and follow-up time of the patients were 20 (23.6) hours, 9 (4.3) days, and 12 (25.9) months, respectively. The mean (SD) of blood oxygen saturation was 85 (16.9) percent after surgery. The criterion for operative success in our study was a difference in oxygen saturation pre-operatively vs. post-operatively of greater than 10%. In this regard, the operation was successful in 134 (77.9%) cases. The overall morbidity was 15 out of 180 (8.7%) patients. Death occurred in 12 cases after surgery, so the mortality risk was 6.7%.

Success of operation

Table I shows the rate of operative success based on the underlying disease.

Table I. Operative success based on underlying disease

	Unsuccessful	Successful	Success Rate
TF	14	53	79.1%
PA	6	16	72.7%
CTPS	7	30	81.1%
TA	2	15	88.2%
DORV&PS	3	11	78.5%
SV&PS	7	16	69.6%
Total	39	141	77.9%

TF = Tetralogy of Fallot, PA = pulmonary atresia, CTPS = complete transposition with pulmonary stenosis, TA = tricuspid atresia, DORV & PS = double outlet right ventricle with severe pulmonary stenosis (DORV & PS), SV & PS = single ventricle with pulmonary stenosis

Crude associations between the predictor variables were assessed by comparing their values between the patients who had successful vs. unsuccessful surgery. The results are presented in Table II.

The mean O₂ saturation level was significantly lower in patients with successful operation ($P < 0.001$). The other variables did not show any significant results.

Also, the logistic regression analysis showed that there was a significant reverse adjusted

association between this predictor and success of surgery ($\beta = -0.20$, $P < 0.001$; OR = 0.82 [CI 95%: 0.76 - 0.88]).

Table II. Comparison of data between patients with or without successful results of surgery

	Success (n = 134)	No success (n = 38)	P-value
Age (mo)	30 (42)	24 (58.5)	0.75
Sex			0.20
Male	71 (53%)	24 (63.2%)	
Female	63 (47%)	14 (36.8%)	
Weight kg	11 (9.2)	10.5 (12.5)	0.57
O ₂ saturation before surgery (%)	76.3 (8.6)	58.4 (13.8)	< 0.001
Underlying disease			0.61
TF	51 (38.1%)	14 (36.8%)	
PA	15 (11.2%)	5 (13.2%)	
CTPS	30 (22.4%)	6 (15.8%)	
TA	13 (9.7%)	2 (5.3%)	
DORV & PS	10 (7.5%)	3 (7.9%)	
SV & PS	15 (11.2%)	8 (21.1%)	
Pulmonary artery diameter			0.34
Small	28 (20.9%)	3 (7.9%)	
Medium	73 (54.5%)	26 (68.4%)	
Normal	33 (24.6%)	9 (23.7%)	
Status of operation			0.61
Elective	128 (95.5%)	37 (97.4%)	
Urgent	6 (4.5%)	1 (2.6%)	
Technique of Operation			0.55
MBTS	121 (90.3%)	34 (89.5%)	
CBTS	3 (2.2%)	0	
CS	10 (7.5%)	4 (10.5%)	
Surgical incision			0.51
Right Thoracotomy	87 (64.9%)	22 (57.9%)	
Left Thoracotomy	37 (27.6%)	11 (28.9%)	
Mid-sternotomy	10 (7.5%)	5 (13.2%)	

TF = Tetralogy of Fallot, PA = pulmonary atresia, CTPS = complete transposition with pulmonary stenosis, TA = tricuspid atresia, DORV & PS = double outlet right ventricle with severe pulmonary stenosis (DORV&PS), SV & PS = single ventricle with pulmonary stenosis, MBTS = modified Blalock-Taussig shunt, CBTS = classic Blalock-Taussig shunt, CS = central shunt

Morbidity

There were 15 (8.7%) cases of post-operative complications. The most common complication was post-operative bleeding leading to re-exploration, which occurred in 5 patients. Fig. 1 shows the different causes of post-operative morbidities among the patients.

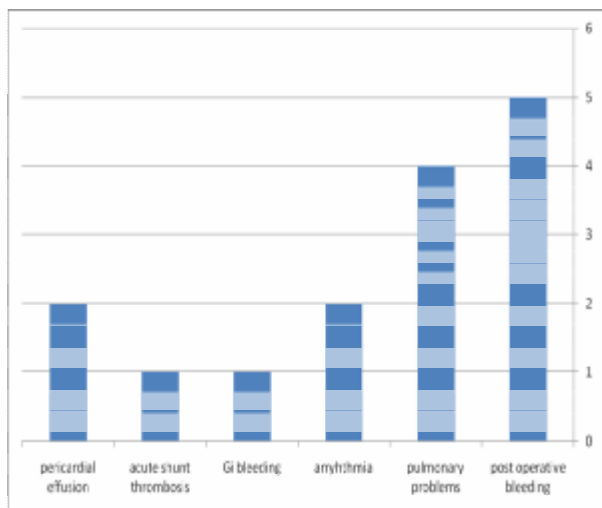


Fig. 1. Frequency of different causes of morbidity after surgery among patients

The results of the bivariate analysis are presented in Table III.

It was suggested that the male sex could be a risk factor for overall morbidity ($P = 0.04$; $OR = 3.66$ [CI 95%: 0.96 – 13.66]). Also, it was observed that the MBTS technique was performed more often in the patients without any morbidity than in those with morbidity. The other variables did not show any significant difference.

The multivariate analysis revealed that none of the above-mentioned variables could predict the overall morbidity. The male sex showed only a borderline significant result ($\beta = 1.20$ [CI 95%: -0.08-2.50]; the Wald test $P = 0.07$).

Mortality

There were 12 (6.7%) deaths within 30 days of the procedure, which were divided into cardiac and non-cardiac deaths.

The 5 (2.77%) cases of cardiac-related death included:¹ A 4-year-old boy with pulmonary atresia, who underwent classic shunt.

After surgery, refractory ventricular fibrillation developed, leading to death.² A 7-year-old girl with pulmonary atresia, who underwent emergency operation due to hypoxic spells, died in the immediate post-operative period of sudden cardiac arrest and unsuccessful resuscitation.³

Table III. Comparison of data according to morbidity of patients after operation

	Morbidity (n = 15)	No morbidity (n = 157)	P-value
Age (mo)	48 (42)	24 (46.7)	0.36
Sex			0.04
Male	12 (80%)	82 (52.2%)	
Female	3 (20%)	75 (47.8%)	
Weight (kg)	15 (7.8)	11 (10.3)	0.15
O₂ saturation before surgery (%)	61 (12)	62 (15.1)	0.77
Underlying diseases			0.63
TF	4 (26.7%)	61 (38.9%)	
PA	1 (6.7%)	19 (12.1%)	
CTPS	4 (26.7%)	32 (20.4%)	
TA	1 (6.7%)	14 (8.9%)	
DORV & PS	1 (6.7%)	12 (7.6%)	
SV & PS	4 (26.7%)	19 (12.1%)	
Pulmonary artery diameter			0.26
Small	6 (40%)	26 (16.6%)	
Medium	5 (33.3%)	93 (59.2%)	
Normal size	4 (26.7%)	38 (24.2%)	
Urgency of operation			0.90
Elective	14 (93.3%)	150 (95.5%)	
Urgent	1 (6.7%)	7 (4.5%)	
Technique of Operation			0.08
MBTS	11 (73.3%)	143 (91.1%)	
CBTS	1 (6.7%)	2 (1.3%)	
CS	3 (20%)	12 (7.6%)	
Surgical incision			0.30
Right Thoracotomy	9 (60%)	99 (63.1%)	
Left Thoracotomy	3 (20%)	45 (28.7%)	
Mid-sternotomy	3 (20%)	13 (8.2%)	

TF = Tetralogy of Fallot, PA = pulmonary atresia, CTPS = complete transposition with pulmonary stenosis, TA = tricuspid atresia, DORV & PS = double outlet right ventricle with severe pulmonary stenosis (DORV & PS), SV & PS = single ventricle with pulmonary stenosis, MBTS = modified Blalock-Taussig shunt, CBTS = classic Blalock-Taussig shunt, CS = central shunt

A 2-month-old infant with pulmonary atresia, who underwent MBTS, developed heart failure without response to resuscitation and expired.⁴ A 1-month-old neonate with

tricuspid atresia and right ventricular hypoplasia in hypoxic spell underwent emergency MBTS but died in the operating room due to refractory ventricular fibrillation.⁵ A 6-month-old boy with pulmonary atresia died in the immediate post-operative period of sudden cardiac arrest and unsuccessful resuscitation.

Non-cardiac deaths were due to respiratory failure (4 cases), sepsis (2 cases), and massive gastrointestinal bleeding (1 case). Most of these non-cardiac deaths were in the early period of our study, and the rate of these mortalities decreased with time. The bivariate analysis showed that there were associations between mortality and age ($P = 0.009$), weight ($P = 0.003$), pulmonary artery diameter ($P = 0.029$), and urgency of operation ($P < 0.001$; OR= 32.4 [CI 95%: 8.2 – 128.1]). The results are presented in Table IV.

Table IV. Comparison of data according to mortality of patients after operation

	Dead (n = 12)	Alive (n = 168)	P-value
Age (mo)	4 (27.3)	24 (46.5)	0.009
Sex			0.77
Male	6 (50%)	91 (54.4%)	
Female	6 (50%)	77 (45.6%)	
Weight (kg)	5.5 (5)	11 (10.1)	0.003
O ₂ saturation before surgery (%)	58.6 (11.6)	62.5 (15)	0.37
Underlying diseases			0.18
TF	4 (33.3%)	62 (37.3%)	
PA	4 (33.3%)	19 (11.2%)	
CTPS	1 (8.3%)	36 (21.3%)	
TA	2 (16.8%)	15 (8.9%)	
DORV & PS	1 (8.3%)	13 (7.7%)	
SV & PS	0	23 (13.6%)	
Pulmonary artery diameter			0.03
Small	4 (33.3%)	28 (16.6%)	
Medium	8 (66.7%)	98 (58.6%)	
Normal size	0	42 (24.8%)	
Urgency of operation			< 0.001
Elective	5 (41.7%)	161 (95.9%)	
Urgent	7 (58.3%)	7 (4.1%)	
Technique of Operation			0.10
MBTS	9 (75%)	152 (90.5%)	
CBTS	1 (8.3%)	2 (1.2%)	
CS	2 (16.7%)	14 (8.3%)	
Surgical incision			0.09
Right Thoracotomy	10 (83.3%)	105 (62.7%)	
Left Thoracotomy	0	48 (28.4%)	
Mid-sternotomy	2 (16.7%)	15 (8.9%)	

TF = Tetralogy of Fallot, PA = pulmonary atresia, CTPS = complete transposition with pulmonary stenosis, TA = tricuspid atresia, DORV & PS = double outlet right ventricle with severe pulmonary stenosis (DORV & PS), SV & PS = single ventricle with pulmonary stenosis, MBTS = modified Blalock-Taussig shunt, CBTS = classic Blalock-Taussig shunt, CS = central shunt

In the multivariate analysis, a Poisson regression model was fitted. Because of the complexity of the model and the lack of any significant association in the bivariate analysis, underlying diseases were not considered in the model. The presence of urgent situations for surgery had a significant association with the occurrence of mortality ($\beta = 2.45$ [CI 95%: 1.11- 3.88]; the Wald test $P < 0.001$). Also, weight showed a significant reverse association with mortality ($\beta = -0.34$ [CI 95%: -0.64 - -0.030]; the Wald test $P = 0.003$). The other variables had no important correlations.

Discussion

Systemic-pulmonary arterial shunts were developed in 1945 by Blalock and Taussig and later by Potts and Waterston.^{10,12-14} The basis of this operation was increasing the pulmonary blood flow in order to decrease cyanosis in cyanotic heart diseases.

Multiple studies about the factors that influence the results of systemic-pulmonary shunt have been performed.¹⁵⁻¹⁷

Tsai and co-workers from January 1986 to December 1991 performed 100 modified Blalock-Taussig shunts, using PTFE grafts on 86 patients with complex cyanotic congenital cardiac malformations. The patients' ages ranged from 15 days to 22 years. The mean follow-up period was 13.5 months (range, one - 50 months). There was one hospital mortality and one patient with morbidity. The univariate analysis showed the age of the patients operated upon and the graft size were the risk factors for shunt patency.¹⁴

Alkhulaifi and co-workers performed 79 systemic-pulmonary shunts in 75 neonates with cyanosis and severely reduced pulmonary blood flow between March 1993 and December 1998. The mean age was 11.5 days, and the mean weight was 3 kg; the 30-day mortality was 3 (4%) patients. The univariate and logistic regression analyses revealed a weight less than 2 kg ($P=0.039$) and pre-operative mechanical ventilation

($P=0.008$) to be the predictors of early mortality, whereas pulmonary hypoplasia ($P=0.55$), diagnostic group ($P=0.79$), shunt size ($P=0.2$), and surgical approach ($P=0.5$) were not. They concluded that the systemic-pulmonary shunt remained an effective palliative measure in cyanotic neonates despite specific complications.⁷

Our study evaluated 180 patients who had undergone systemic-pulmonary shunt between 1992 and 2006. All of the patients had cyanotic heart disease and needed systemic-pulmonary shunt. One surgical group performed all the operations, and anesthesia and post-operative care were similar in all the patients.

The mean age and weight of our patients were 44.5 months and 13.23 kilograms, respectively. The most common technique utilized was modified Blalock - Taussig shunt (90%) through a right thoracotomy (64.4%). The pulmonary artery diameter was small (z value < -2) in 17.8% of the patients. Almost eight percent of the operations were performed in an emergency setting. The mean partial pressure of oxygen (PO_2) and O_2 saturation rise was 24.35 mmHg and 25.31%, respectively. We found 77.8% success, 6.7% mortality, and 8.7% morbidity rates.

In order to determine the risk factors of unfavorable operative outcome, we evaluated different variables. They included age, sex, and weight of the patients, underlying diseases, surgical incision and technique, pulmonary artery diameter, urgency of operation, and pre-operative O_2 saturation.

Among these factors, none of them had a significant influence on the success rate of the operation. In our results, the morbidity rate was higher in the male patients with small pulmonary artery diameters (z value < -2), longer duration of ICU stay, and in those who were under mechanical ventilation pre-operatively because of critical low O_2 saturation. The multivariable analysis, however, did not confirm any predictive factor for operative morbidity except for a weak influence of the male sex.

The bivariate analysis showed that there were associations between mortality and age, weight, pulmonary artery diameter, and urgency of operation; however, after the multivariate analysis, only low body weight (less than 6 kilograms) and urgency of operation had a significant association with the occurrence of mortality.

It should be mentioned that there are some limitations in our study. The retrospective nature of the study makes an evaluation of some aspects of the problem impossible. On the other hand, the past fifteen years have seen many great developments in cardiac surgery practice, which have surely influenced the surgical results. We, however, could not enter all of them in the study. Finally, the loss of follow-up of about 16% might be a weak point in the evaluation of the results.

In conclusion, systemic-pulmonary shunt remains an effective palliative measure in cyanotic heart disease with high success rates and low complications. Performing the operation in low weight infants and in the emergent setting is the important challenge. Obviously, this may be handled with proper surgical principles and a cooperative health care team.

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Conflict of Interest

No conflicts of interest have been claimed by the authors.

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