“Reverse Blalock-Taussig shunt”: Application in single ventricle hybrid palliation

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Objective: Retrograde aortic arch malperfusion after ductal stenting can be life-threatening after univentricular hybrid palliation. Arch perfusion can be maintained with a main pulmonary artery to innominate artery shunt placed during the stage I procedure: a “reverse Blalock-Taussig shunt.”

Methods: A retrospective review of 37 infants who underwent hybrid palliation from January 2004 to March 2010 was performed. The infants were divided into 2 groups, those with (group I, n = 16) and those without (group II, n = 21) a reverse Blalock-Taussig shunt.

Results: At the initial palliation, no differences were found in the demographics, systolic or diastolic pressures, or ventricular or atrioventricular valve function between the 2 groups. Group I had more infants with aortic atresia ($P < .01$) and smaller ascending aortas ($P < .01$). Before stage II, the retrograde aortic Doppler flow velocity increased in group I ($P < .01$) and was unchanged in group II. The reintervention rates before stage II were similar between the 2 groups. Before stage II, the ventricular end-diastolic pressure, left and right pulmonary artery pressures and diameters, and mixed venous and arterial saturations were similar between the 2 groups. The complication rates between the 2 groups were not significantly different, although a nonsignificant trend toward more neurologic complications was noted in group I. The Kaplan-Meier survival estimate at 1 year was similar between the 2 groups (63% for group I vs 71% for group II).

Conclusions: The presence of a reverse Blalock-Taussig shunt was not associated with more adverse events than those without. Gradual retrograde arch obstruction occurs commonly in palliated infants with aortic atresia. A reverse Blalock-Taussig shunt might play an important role to address the potential of retrograde obstruction, augmenting arch blood flow. (J Thorac Cardiovasc Surg 2013;146:352-7)

The so-called hybrid palliation (bilateral pulmonary artery [PA] bands and a ductal stent) has emerged as an alternative to Norwood palliation for infants with hypoplastic left heart syndrome or its variants.¹⁻³ The potential, but unproved, advantage of hybrid palliation is the avoidance of circulatory bypass and aortic arch reconstruction in the neonatal period, shifting such procedures to later in life. A potential problem with this strategy is either immediate or delayed obstruction in the aortic isthmus after ductal stent deployment. This obstruction can compromise retrograde aortic arch flow, which supports, in the extreme case of aortic valvar atresia, the cerebral and coronary

perfusion. A potential solution is the placement of a main PA to innominate artery shunt, analogous to a Blalock-Taussig (BT) shunt, which we have designated the “reverse BT (revBT) shunt.”⁴ This connection can provide a source of blood flow to the ascending aortic arch if the retrograde arch flow from the isthmus becomes compromised. Few centers perform a revBT shunt during hybrid palliation, and little is known regarding the outcomes and potential advantages and disadvantages of the procedure. In the present study, we reviewed our experience during hybrid palliation in those infants with and without a revBT shunt.

METHODS

A retrospective chart review was conducted of all infants with hypoplastic left heart syndrome or 1 of its variants, who had undergone hybrid single ventricle palliation from January 2004 to March 2010. The Research Ethics Board at the Hospital for Sick Children approved the study, and patient and family consent was waived owing to the retrospective nature of the study. The form of surgical palliation (hybrid [with or without a revBT shunt], Norwood, or primary transplantation) was discussed at an interdisciplinary conference after presenting the options to the family. No specific decision-making protocol was applied for the form of surgical palliation. The revBT shunt was placed after PA banding, avoiding the pulmonary valve ring to the innominate artery using a 3.5-mm polytetrafluoroethylene graft. The use of the revBT shunt was approved by the Surgical Innovations Ethics Committee, and the experimental nature was explained to each family.³

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Abbreviations and Acronyms

AVVR	=	atrioventricular valve regurgitation
BT	=	Blalock-Taussig
ECMO	=	extracorporeal membrane oxygenation
PA	=	pulmonary artery
PVR	=	pulmonary valve regurgitation
revBT	=	reverse BT

Echocardiographic Assessment

Echocardiography was performed using a Phillips ATL (Advanced Technology Laboratories, Bothell, Wash), Philips IE-33 (Philips Medical systems, Eindhoven, The Netherlands), or Vivid 7 (GE Healthcare, Waukesha, Wis) system. The echocardiograms at diagnosis, immediately after hybrid palliation, before hospital discharge, and before the comprehensive stage II procedure (arch reconstruction and a cavopulmonary connection) were reviewed ($n = 27$). Digital images were stored and analyzed offline with commercially available software (SyngoDynamics, Siemens Medical Systems, Erlangen, Germany). Ventricular function was graded visually (1, normal; 2, mild impairment; 3, moderate; and 4, severe impairment), and the degree of atrioventricular valve regurgitation (AVVR) and pulmonary valve regurgitation (PVR) was graded (1, none/trace; 2, mild; 3, moderate; and 4, severe) according to the color flow regurgitant jet, as previously described.^{5,6} From a suprasternal long-axis view, isthmus flow was measured at the base of the left subclavian artery. The flow profile was outlined manually to measure the maximal flow velocity, and the velocity–time integral, with 3 consecutive heart beats, was measured and averaged. The retrograde/antegrade velocity–time integral ratio was calculated and considered dominantly retrograde if the ratio was >1 . The presence of flow in the revBT shunt was determined; however, because of the incident angle, the flow velocity could not be assessed.

Catheterization and Angiography

The invasive hemodynamic studies and angiograms before stage II surgery were reviewed. The diameters of the right and left PAs were measured at the hilum, proximal to the takeoff of the branching vessels. A Nakata index was calculated as the sum of the right and left PA cross-sectional areas indexed to the child's body surface area.⁷

Statistical Analysis

The data are presented as the mean $\pm$ standard deviation. Continuous variables were compared using the Mann-Whitney U test and Student t test. Dichotomous and categorical variables were analyzed using Fisher's exact test and the chi-square test. Paired data were examined using paired 2-tailed t tests. Kaplan-Meier curves were constructed to determine the freedom from death or transplantation, and the survival rate was compared between the 2 groups using a log-rank test.

RESULTS

Patient Characteristics

A total of 48 consecutive neonates underwent hybrid palliation. Excluded from the present review were those neonates who underwent hybrid palliation as a bridge to transplantation ($n = 7$), a salvage procedure during extracorporeal membrane oxygenation (ECMO) support ($n = 1$) or as an interim procedure before biventricular repair ($n = 3$). The remaining 37 neonates included in the study cohort were divided into 2 groups: those with (group I) and

those without (group II) a revBT shunt. The patient characteristics and diagnoses are listed in Table 1. In group I, 16 neonates underwent palliation at a median age of 6.5 days (mean, 7.4 ± 4.8 days) and median weight of 3.3 kg (mean, 3.2 ± 0.6 kg). The 21 neonates in group II underwent hybrid palliation without a revBT shunt at a median age of 7.0 days (mean, 13.5 ± 24 days) and median weight of 3.2 kg (mean, 3.2 ± 0.5 days). In group II, 4 neonates had aortic atresia; 2 procedures were abandoned because of patient instability during the procedure (electrocardiographic changes of ischemia). The other 2 neonates had an ascending aorta more than 3 mm in diameter and presented early in our experience before the decision to place a revBT shunt in neonates with aortic atresia. No demographic differences were seen between the 2 groups.

Echocardiographic Findings

Not unexpectedly, the group I infants had a significantly smaller ascending aorta (2.4 ± 1.2 mm vs 5.0 ± 1.9 mm, $P < .01$) and more commonly aortic atresia (16/16 [100%] vs 4/21 [19%]; $P < .01$) than those in group II (Table 2). Although all neonates in group I by anatomic definition had retrograde arch flow, 11 neonates (52%) in group II had a dominant retrograde arch flow pattern. No significant difference was found in ventricular function or the degree of AVVR or PVR at the initial examination between the 2 groups.

Interstage Echocardiography: After Stage I and Before Stage II

Echocardiographic studies were obtained before stage II at 186 ± 24 and 198 ± 38 days of age in group I and II, respectively (Table 3). All revBT shunts were patent, as determined by echocardiography or angiography. No significant differences were found in ventricular function, AVVR, or PVR during this period in either group. Before stage I, no difference was found in retrograde flow between the 2 groups; however, in group I, the retrograde arch flow peak velocities increased significantly ($P < .01$). In group II (those with dominant retrograde arch flow, $n = 11$), no significant increase was seen in the velocity ($P = .30$). In the 3 group II infants (19%) with aortic atresia, the changes in retrograde peak flow velocities were as follows: patient 1, from 0.95 to 2.2 m/s; patient 2, 1.5 to 2.6 m/s; and patient 3, 1.7 to 2.4 m/s. Although the analysis of the entire group showed no statistically significant difference, a clear trend was seen toward progressive arch malperfusion in these infants.

Subsequent Interventions

Intervention on the atrial septum was performed in 10 of 16 (63%) infants in group I and 11 of 21 infants (52%) in group II. No significant difference was seen in the age at intervention between the 2 groups (group I, 52 ± 52 days;

TABLE 1. Patient characteristics and diagnosis

Variable	Group I (n = 16)	Group II (n = 21)	P value
Male gender (%)	8 (50)	7 (33)	.31
Antenatal diagnosis (%)	9 (56)	16 (76)	.20
Age at stage I (d)	7.4 ± 4.8	13.5 ± 24	.46
Weight at stage I (kg)	3.2 ± 0.6	3.2 ± 0.5	.94
Hypoplastic left heart syndrome			—
AA/MA	12	3	
AA/MS	3	1	
AS/MA		1	
AS/MS		7	
TA, TGA	1	1	
TAPVD*	1		
Unbalanced AVSD		5	
DORV, CoA		2	
DILV, TGA		1	

AA, Aortic atresia; MA, mitral atresia; MS, mitral stenosis; AS, aortic stenosis; TA, tricuspid atresia; TGA, transposition of great arteries; TAPVD, total anomalous pulmonary venous drainage; AVSD, atrioventricular septal defect; DORV, double-outlet right ventricle; CoA, coarctation of aorta; DILV, double-inlet left ventricle. *Patient with TAPVD had underlying diagnosis of AA/MA.

median, 42 days; vs group II, 58 ± 34 days; median, 57 days; $P = .32$). An intervention on the ductal stent was performed in 3 of 16 infants (19%) in group I and 2 of 21 (10%) in group II at 12, 49, and 123 days of age in group I and 13 and 14 days of age in group II. One child in group I underwent coil embolization of an aortopulmonary collateral 4 months before the comprehensive stage II procedure. The intervention rates before stage II were not different between the 2 groups ($P = .21$).

Growth, PA Size, and Hemodynamic Findings Before Stage II

Somatic growth was not significantly different nor was the size of the branch PAs, with Nakata indexes also similar

TABLE 2. Preoperative echocardiographic findings

Variable	Group I (n = 16)	Group II (n = 21)	P value
Before stage I			
Patients (n)	16	21	
Ascending aorta size (mm)	2.4 ± 1.2	5.0 ± 1.9	<.01
Aortic atresia (n)	16 (100%)	4 (19%)	<.01
Retrograde arch flow (n)	16 (100%)	11 (52%)	<.01
Ventricular function grade	1.3 ± 0.7	1.1 ± 0.5	.42
AVVR	1.4 ± 0.5	1.2 ± 0.8	.53
PVR	1.2 ± 0.4	1.1 ± 0.2	.18
Before stage II			
Patients (n)	11	16	
Ventricular function grade	1.1 ± 0.3	1.1 ± 0.3	.87
AVVR	1.6 ± 0.5	2.0 ± 0.5	.19
PVR	1.4 ± 0.5	1.3 ± 0.5	.55
Retrograde arch flow (m/s)	1.9 ± 0.4	1.8 ± 0.6*	.29

AVVR, Atrioventricular valve regurgitation; PVR, pulmonary valve regurgitation.

*Nine patients had dominant retrograde flow.

TABLE 3. Interstage echocardiographic findings

Group	After stage I	Before stage II	P value
I (n = 11)			
Age (d)	26 ± 9.6	186 ± 24	—
Ventricular function grade	1.2 ± 0.4	1.1 ± 0.3	.64
AVVR	2.0 ± 0.4	1.6 ± 0.5	.09
PVR	1.5 ± 0.5	1.4 ± 0.5	.67
Retrograde arch flow (m/s)	1.2 ± 0.2	1.9 ± 0.4	<.01
II (n = 16)			
Age (d)	26 ± 16	198 ± 38	—
Ventricular function grade	1.0 ± 0.0	1.1 ± 0.3	.69
AVVR	1.8 ± 0.5	2.0 ± 0.5	.28
PVR	1.1 ± 0.3	1.3 ± 0.5	.59
Retrograde arch flow (m/s)*	1.6 ± 0.5	1.8 ± 0.6	.3

AVVR, Atrioventricular valve regurgitation; PVR, pulmonary valve regurgitation.

*Nine patients had dominant retrograde flow.

between the 2 groups (Table 4). The hemodynamic outcomes before stage II were not different between the 2 groups (Table 4). In particular, no difference were found in diastolic blood pressure, mixed venous saturation, or chamber end-diastolic pressure.

Complications

In group I, 10 complications occurred in 7 children. One neonate developed hypotension after insertion of the revBT shunt, and the shunt was clipped, with recovery of blood pressure. This same child developed necrotizing enterocolitis the following day. Another child developed recurrent atrial flutter requiring cardioversion during the procedure and experienced bilateral, watershed cerebral infarctions. Eight additional complications occurred after the index hybrid procedure, including atrial flutter and migration of an atrial stent. The atrial stent was deployed 7 days after stage I to address a highly restrictive septal defect; however, the stent embolized, requiring surgical removal. The neurologic

TABLE 4. Catheterization data before stage II

Variable	Group I (n = 11)	Group II (n = 16)	P value
Age (mo)	3.8 ± 0.9	4.2 ± 1.1	.34
Body weight (kg)	5.6 ± 1.0	5.8 ± 1.2	.65
Systolic blood pressure (mm Hg)	83.6 ± 11.7	88.8 ± 8.7	.14
Diastolic blood pressure (mm Hg)	45.6 ± 9.5	46.6 ± 6.9	.54
Ventricular end-diastolic pressure (mm Hg)	7.5 ± 2.6	6.8 ± 2.6	.47
Mean right PA pressure (mm Hg)	10.8 ± 2.8	11.9 ± 3.7	.45
Mean left PA pressure (mm Hg)	13.5 ± 4.5	13.8 ± 5.5	.89
Qp/Qs	0.95 ± 0.4	0.88 ± 0.4	.63
Finger tip oximetry (room air) (%)	76.8 ± 4.9	80.1 ± 6.5	.22
Mixed venous saturation (%)	53.7 ± 9.3	59.7 ± 9.9	.14
Right PA diameter (mm)	8.1 ± 1.3	7.5 ± 1.9	.43
Left PA diameter (mm)	7.1 ± 1.7	6.9 ± 1.7	.77
Nakata index	323 ± 83	292 ± 150	.54

PA, Pulmonary artery; Qp/Qs, pulmonary-to-systemic flow ratio.

FIGURE 1. Outcomes of patients undergoing hybrid palliation with (group I) or without (group II) reverse Blalock-Taussig shunt (*revBT*). *BCPS*, Bidirectional cavopulmonary shunt; *+RBT*, reverse Blalock-Taussig shunt.

complications were as follows: 1 child with combined right parietal and frontal cerebral infarctions, 1 left cerebral hemorrhage (which occurred 2 months after the procedure), and the previously mentioned patient with bilateral watershed infarctions secondary to hypotension. Three children developed necrotizing enterocolitis during the initial hospitalization, 1, 1, and 9 days after stage I.

In group II, 10 complications developed, 7 during the stage I procedure. Two children required ECMO support secondary to hypotension, and two developed ischemic electrocardiographic changes during surgery (with one abandonment of a planned *revBT* shunt). Intraoperative supraventricular tachycardia was noted in 3 neonates. Three complications occurred after stage I, including spontaneous loosening of one left PA band, one right femoral artery thrombosis, and one left diaphragmatic paralysis. No neurologic complications occurred in the group II cohort. No statistically significant difference was found in the complication rates between the 2 groups.

Outcomes

Of the 16 infants in group I and 21 in group II, 11 (69%) and 17 (81%) underwent the stage II procedure. One infant in group II underwent attempted *revBT* shunt placement 2 days after the index hybrid palliation. This infant subsequently required Norwood palliation at 102 days and then progressed to a bidirectional cavopulmonary connection at 9 months of age (290 days after the index hybrid palliation). No statistically significant difference was found in survival to the stage II procedure between the 2 groups ($P = .74$). At the last follow-up visit, 5 children in group I and 8 in group II were awaiting Fontan palliation

and 5 infants in group I and 7 in group II had undergone a Fontan procedure (Figure 1). The Kaplan-Meier curve for freedom from death or transplantation is shown in Figure 2, with no difference in the 1-year death or transplantation-free survival between the 2 groups (group I, 63%, and group II, 76%, log rank $P = .47$). Of the 4 neonates with aortic atresia who did not receive a *revBT* shunt, the 2 with an ascending aorta more than 3 mm in diameter underwent the stage II procedure uneventfully. Of the 2 neonates whose arch was less than 3 mm, 1 died after stage I and the other required ECMO to rehabilitate the myocardium before stage II. All 3 patients were awaiting Fontan palliation at last follow-up.

DISCUSSION

So-called hybrid palliation (PA banding and ductal stenting) has become an alternative to the Norwood procedure. As an evolving palliation strategy, a variety of issues and complications are becoming apparent. One such issue is the management of present or potential retrograde arch malperfusion.⁸ In the hybrid circulation with aortic atresia, coronary and cerebral perfusion is dependent on retrograde arch flow. As such, retrograde arch obstruction will result in coronary and cerebral vascular bed ischemia. The *revBT* shunt was developed to provide an alternative source of upper body blood flow if retrograde arch flow became compromised.⁴ What has not been established is the effect on the clinical course for those neonates who had received a *revBT* shunt.

In the present study cohort, no significant differences were found in the overall stage II and 1-year survival for those with and without a *revBT* shunt. All neonates in group

FIGURE 2. Kaplan-Meier survival analysis comparing groups I and II. Data in parentheses indicate number of patients. No significant difference was found in 1-year survival: group I, 63%; group II, 76%; log-rank $P = .47$.

I had aortic atresia and, subsequently, a smaller ascending aorta than those in group II, and, using a Norwood strategy, such neonates were considered at greater risk.^{9,10} Others have reported that an ascending aorta diameter less than 2 mm is a contraindication for bilateral PA banding.¹¹ From our cohort, the median ascending aorta size in group I was 2 mm, with one half ($n = 8$) having a diameter less than 2 mm. Nonetheless, the outcomes of group I were equivalent to those of group II.

The interstage echocardiographic data revealed that the retrograde arch flow peak velocity in group I increase significantly over time, with no significant changes in group II. These findings support the notion that those infants with aortic atresia are at greater risk of progressive retrograde flow obstruction. The question can be raised of whether the revBT shunt in and of itself increases or potentiates progressive retrograde arch obstruction or whether it is related to the child's underlying anatomic substrate (aortic atresia and severe hypoplasia of the ascending aorta). In this regard, Stoica and colleagues⁸ found that neonates with aortic atresia (with hybrid palliation but no revBT shunt) were more likely to develop retrograde arch obstruction. The interstage echocardiographic data from the present cohort showed the absence of significant differences in ventricular function or degree of AVVR in the 2 groups. These results suggest that coronary perfusion in group I was secured, despite the progress in aortic isthmus obstruction by the presence of the shunt. The rate of unplanned interstage procedures was also not different between the 2 groups. Additionally, no statistically significant differences were found in the presence of PVR before stage II between the 2 groups, suggesting that the presence of the shunt (inserted near the pulmonary valve) was not adversely affected.

Previously published data from our institution and patient cohort have shown that no differences in the right or left PA diameters or Nakata index between infants undergoing either Norwood or hybrid palliation.^{12,13} Similarly, in the present study of hybrid, palliated neonates, no differences were seen in the PA diameters or Nakata indexes between the 2 groups. No demonstrable difference or detrimental effect was found for the revBT shunt on PA growth. One of the greatest concerns with the addition of a revBT shunt is the potential for increased neurologic complications related to the shunt. The incidence of neurologic complications was greater in the revBT shunt group, although it did not reach statistical significance. In 1 neonate with right cerebral infarction, the shunt had been placed on the ipsilateral side and was considered a likely source of thrombus. A second neonate had recurrent atrial flutter and bilateral watershed cerebral infarctions, which were attributed to hypotension and not directly shunt related. The final infant experienced a cerebral hemorrhage on the contralateral side of the shunt 2 months after the procedure. This infant had been treated with low-molecular-weight heparin, which was considered a significant contributing factor to the hemorrhage. In contrast, the prevalence of neurologic deficits persisting at discharge after a Norwood operation has been reported to be 3%.¹⁴ One of the potential advantages of the hybrid strategy is the avoidance of cardiac bypass in the neonatal period, a documented risk factor for cerebral injury.¹⁵ We could not fully determine whether a revBT shunt exposes the infant to a greater risk of cerebral events owing to the small numbers in the present study.

Between stages, strategies to address infants without a revBT shunt who develop isthmus obstruction include

immediate conversion to a Norwood circulation, stent deployment within the stenotic aortic isthmus, an early stage II procedure, and heart transplantation. In the present study, 2 infants in group II underwent a Norwood procedure. The first infant experienced cardiovascular collapse during the hybrid procedure. A revBT shunt was added, and the child was supported with ECMO. A Norwood procedure was electively performed after the cardiac function recovered. A second infant could not undergo a comprehensive stage II procedure because of pulmonary hypertension (due to a loose PA band) and was converted to Norwood circulation.

Stoica and colleagues⁸ reported that stent insertion in the stenotic isthmus has proved to be an effective and durable solution to isthmus obstruction. Once isthmus obstruction has occurred, the risks of sudden death or myocardial failure are much greater. What might be more important than solutions to the address such problems once they have developed are approaches to protect the at-risk patient group, with consideration of the addition of a revBT shunt to modulate such a complication. Our institutional practice is to place a revBT shunt prophylactically as a proactive strategy for those hybrid patients who are dependent on retrograde flow or who are at high risk of progressive obstruction.

Study Limitations

The present study was a retrospective, nonrandomized review of a small cohort from a single institution and thus had the limitations intrinsic to those boundaries. Additionally, the lack of an institutional protocol to allocate neonates to a hybrid strategy with or without a revBT shunt regardless of outflow anatomy could have avoided the possible bias intrinsic to retrospective review.

CONCLUSIONS

A revBT shunt was placed for infants thought to be at greater risk of upper body ischemia from compromised retrograde blood flow. The outcomes were equivalent to those without a revBT shunt. Although the retrograde arch flow velocity significantly increased in the group with a revBT shunt, ventricular function and AVVR were preserved. The cohort with a revBT shunt had comparable hemodynamic findings, with no detrimental effects on PA growth. The rates of unplanned interstage procedures and

complications were not different between the 2 groups. A revBT shunt showed a trend toward more neurologic complications; however, the difference did not reach statistical significance. Our findings have shown that a revBT shunt can play an important role in hybrid palliation to address the issue of potential retrograde arch obstruction.

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