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# Outcomes After Bidirectional Cavopulmonary Shunt in Infants Less Than 6 Months Old

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*Objectives.* **We sought to assess the results after bidirectional cavopulmonary shunt (BCPS) in infants <6 months old and to identify risk factors for poor outcome.**

*Background.* **Although BCPS is a well established procedure for the palliation of patients with a single-ventricle heart, there have been very few reports of outcomes after BCPS in young infants.**

*Methods.* **Since 1990, 42 infants between 0.8 and 6.0 months of age (mean [±SD] 3.7 ± 1.4) have undergone BCPS for primary (n = 16) or secondary (n = 26) palliation of tricuspid atresia (n = 13), hypoplastic left heart syndrome (n = 10) or other forms of functional single-ventricle heart (n = 19). Accessory pulmonary blood flow was included in 18 patients. Preoperative and perioperative data were gathered on retrospective review of patient records, and follow-up was conducted by means of direct physician contact or record review.**

*Results.* **The overall hospital mortality rate, including that associated with reoperations, was 4.8% (2 of 42 patients). Seven patients (17%) required reoperation related to the BCPS or pulmonary blood flow in the early postoperative period: Procedures included take-down of the BCPS in four patients, with one early death, and procedures to decrease pulmonary blood flow in three patients. Age <1 month correlated significantly with early death and with early failure of the BCPS (death or take-down). Follow-up of the 37 patients discharged with intact BCPS was obtained at a mean ± SD of 14.3 ± 11.3 months postoperatively, during which time three patients died (at 6.5 ± 2.5 months). The 2-year actuarial survival rate for patients undergoing BCPS at <6 months of age was 86%. Overall freedom from death or take-down (including early and late events) was significantly lower in patients <2 months old than in those >2 months old. Four patients have undergone successful Fontan completion (18.3 ± 2.9 months postoperatively), and one patient whose BCPS was taken down subsequently underwent successful restoration of a BCPS.**

*Conclusions.* **Outcomes after BCPS in young infants are comparable to those in older infants and children. However, our current preference is to defer this procedure until after 2 months of age.**

**(J Am Coll Cardiol 1997;29:1365–70)**

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The bidirectional cavopulmonary shunt (BCPS) has become a well established procedure for the palliation of functional univentricular hearts (1–7). Its advantages as a primary or second-stage palliative procedure include relief of the volume load on a single functional ventricle, improvement in atrioventricular valve regurgitation, avoidance of pulmonary artery distortion after pulmonary artery banding or systemic to pulmonary artery shunts and prevention of the possible pulmonary vascular obstructive disease that can develop with prolonged ventricular or arterial level left to right shunting. Despite these advantages, there have been few reports of its application in young infants (8,9). The tendency not to perform BCPS in young infants is likely due to a combination of factors, including a history of unfavorable results after classic Glenn shunt in infants (10–12) and uncertainty regarding the reactivity of the pulmonary vasculature in this group of patients.

We routinely perform BCPS at 3 to 6 months of age, after first-stage palliation, in almost all patients with tricuspid atresia, hypoplastic left heart syndrome and other forms of a functional single-ventricle heart. In selected patients with balanced pulmonary blood flow who do not require neonatal intervention, a primary BCPS is performed between 1 and 4 months of age (9). In the present report, we describe our experience with 42 patients <6 months of age who underwent BCPS at our institution between January 1990 and December 1995.

## Methods

**Patients.** Between January 1990 and December 1995, BCPS was performed in 118 patients, 42 (36%) of whom were <6 months of age at the time of the operation. These 42 patients constitute the study group for the present report. Mean age at BCPS was 3.7 ± 1.4 months (range 0.8 to 6.0), and median weight was 5.2 ± 0.9 kg (range 3.5 to 7.5). Figure 1 shows the age distribution of the patients. Primary BCPS was performed in 16 patients (38%), whereas the remaining 26

From the Divisions of Cardiothoracic Surgery and Pediatric Cardiology, University of California San Francisco, San Francisco, California. This study was presented in part at the 45th Annual Scientific Session of the American College of Cardiology, Orlando, Florida, March 1996.

All editorial decisions for this article, including selection of referees, were made by a Guest Editor. This policy applies to all articles with authors from the University of California San Francisco.

Manuscript received July 26, 1996; revised manuscript received January 26, 1997, accepted February 13, 1997.

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Published by Elsevier Science Inc.
0735-1097/97/$17.00
PII S0735-1097(97)00068-5

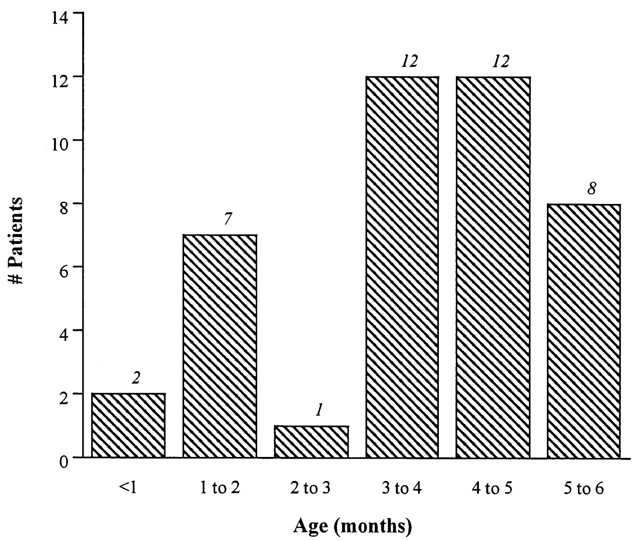


**Figure 1.** Age breakdown of 42 patients undergoing bidirectional cavopulmonary shunt at <6 months old.

patients had undergone one previous palliative procedure. Patient diagnoses are summarized in Table 1.

**Preoperative evaluation.** Preoperative catheterization was performed in 38 patients (91%). Mean arterial oxygen saturation (n = 35) was 76.8 ± 8.2% (range 50% to 94%). Mean pulmonary arterial pressure (n = 28) was 15.8 ± 10.0 mm Hg (range 7 to 49) and mean ventricular end-diastolic pressure (n = 35) was 7.2 ± 2.3 mm Hg (range 3 to 13). Mean indexed pulmonary vascular resistance (n = 27) was 2.2 ± 1.1 Wood units (range 0.6 to 5.9). Indexed pulmonary blood flow (n = 21) averaged 5.4 ± 4.7 ml/min per $m^2$ (range 2.2 to 19.0), and the mean pulmonary to systemic blood flow ratio (n = 21) was 2.0 ± 1.3 (range 0.9 to 6.1). Preoperative echocardiographic evaluation was performed in all patients by an observer who had no knowledge of outcomes or the present study, and 15 patients were found to have mild (n = 10, 24%) or moderate (n = 5, 12%) atrioventricular valve regurgitation.

**Operative procedures.** Thirty-two patients underwent unilateral right (n = 31) or left (n = 1) BCPS, whereas bilateral BCPS was performed in all 10 patients with bilateral superior venae cava. Pulmonary artery augmentation was performed in 11 patients, and seven patients underwent atrial septectomy. Accessory pulmonary blood flow was placed (n = 4) or allowed to remain (from the native anatomy or a previous procedure, n = 14) in 18 patients (43%), as either anterograde flow through a banded or stenotic main pulmonary artery (n = 9) or a systemic to pulmonary artery shunt (n = 9). Additional procedures performed are shown in Table 2. Cardiopulmonary bypass was used in 30 patients, whereas a venous bypass shunt was used in the other 12 patients.

**Table 1.** Patient Diagnoses by Primary Bidirectional Cavopulmonary Shunt and Mean Age

| Diagnosis | All Pts | Pts With Primary BCPS | Age, All Pts (days) |
|---|---|---|---|
| Tricuspid atresia | 13 | 7 | 88.6 ± 47.2* |
| Hypoplastic left heart syndrome | 10 | 0 | 124.3 ± 29.7 |
| Other single ventricle | 19 | 9 | 129.1 ± 30.5 |
| Heterotaxy single ventricle | 7 | 4 | |
| Double-inlet left ventricle | 5 | 2 | |
| Pulmonary atresia with intact ventricular septum | 2 | 0 | |
| Ebstein's anomaly | 1 | 0 | |
| Other | 4 | 3 | |

*Significantly younger than patients with diagnoses other than tricuspid atresia ($p < 0.02$). Data presented are mean value ± SD or number of patients. BCPS = bidirectional cavopulmonary shunt; Pts = patients.

**Table 2.** Additional Procedures Performed at Bidirectional Cavopulmonary Shunt

| Procedure | No. (%) of Pts |
|---|---|
| Systemic to pulmonary artery shunt take-down | 16 (38) |
| Pulmonary artery augmentation | 11 (26) |
| Main pulmonary artery ligation | 9 (21) |
| Atrial septectomy | 7 (17) |
| Pulmonary artery banding | 7 (17) |
| Patent ductus arteriosus ligation | 6 (14) |
| Systemic to pulmonary artery shunt placement | 2 (5) |
| TAPVD repair | 2 (5) |
| Repair stenotic origin of right subclavian artery | 1 (2) |
| Aortic arch augmentation | 1 (2) |
| Mitral valve repair | 1 (2) |
| Aortopulmonary collateral artery ligation | 1 (2) |
| Right ventricle to coronary artery fistula ligation | 1 (2) |

Pts = patients; TAPVD = total anomalous pulmonary venous drainage.

**Data collection and statistical analysis.** Retrospective review of patient records was conducted to gather preoperative and perioperative data. Follow-up was carried out in January 1996 by means of physician contact or direct review of patient records and was complete in all patients. Patients were followed until their most recent physician contact before the time follow-up was conducted, or until they underwent Fontan completion. Data are expressed as either mean value ± SD or as median and range. Preoperative and follow-up arterial oxygen saturations were compared using paired two-tailed *t* test analysis. The Fisher exact test and chi-square test were used to compare dichotomous variables. Logistic regression was used to analyze continuous independent variables for correlation with categoric outcome measures, and was used in forward stepwise fashion for multivariate analysis of variables found to be significant by univariate analysis. Cox proportional hazards regression and Kaplan-Meier product limit methods were used for actuarial survival analysis. Early outcome measures included death and BCPS failure (death or BCPS take-down). Actuarial survival analysis was performed for hospital survivors and for all patients (including those with early events). For the purposes of survival analysis, patients who have undergone Fontan completion were censored at the date of their Fontan procedure. Variables analyzed for correlation with early and late outcomes are listed in the Appendix. SPSS version 6.01 for Windows (SPSS Inc.) was used to perform statistical calculations. Statistical significance was defined as $p \leq 0.05$.

## Results

**Early results.** Patients with tricuspid atresia were significantly younger at BCPS ($p = 0.02$) than patients with other diagnoses. There were two early deaths in the study group (4.8%). One early death was a 61-day old patient with asplenia syndrome who had previously undergone ligation of a patent ductus arteriosus and placement of a systemic to pulmonary artery shunt and who presented for BCPS in extremis, with a pulmonary vascular resistance of 4.7 indexed Wood units, respiratory distress and metabolic acidosis. Because of continued poor oxygenation postoperatively, an additional systemic to pulmonary artery shunt was placed 2 days after BCPS. The patient continued to have low cardiac output and poor oxygen saturation and arrested and died 5 days after BCPS. The other early death was a patient with tricuspid atresia who underwent BCPS at 24 days of age. This patient underwent BCPS take-down to a 3.5-mm systemic to pulmonary artery shunt on the first postoperative day because of persistent low saturations; the patient died later the same day after having a cardiopulmonary arrest.

In addition to this patient, three others underwent BCPS take-down in the early postoperative period. One patient, who underwent primary bilateral BCPS at 30 days of age for palliation of tricuspid atresia with pulmonary stenosis and bilateral superior vena cava, developed presumed viral pneumonia and required extracorporeal membrane oxygenation and BCPS take-down to a systemic to pulmonary artery shunt on postoperative day 3. The BCPS was successfully restored 22 months after the initial failed BCPS in this patient. A patient who underwent primary BCPS at 98 days of age for a double-inlet left ventricle with pulmonary stenosis also manifested a presumed viral pneumonia on the day after surgery, then underwent emergency exploration 2 days later, with reconstruction and banding of a ligated main pulmonary artery to increase pulmonary blood flow. On the fifth postoperative day, the BCPS was taken down to a classic Glenn shunt and the pulmonary artery band was removed. This patient has since undergone main pulmonary artery ligation with placement of a systemic to pulmonary artery shunt. A patient with hypoplasia of the tricuspid valve and right ventricle, along with multiple ventricular septal defects and an atrial septal defect, who underwent primary BCPS at 4.7 months of age, had the BCPS taken down to a systemic to pulmonary artery shunt on an emergency basis 3 days after surgery, after a course complicated by conduction problems and superior vena cava syndrome due to elevated pulmonary artery pressures. Four months later this patient underwent "one and a half ventricle repair," with closure of the ventricular and atrial septal defects, reconstruction of the tricuspid valve and main pulmonary artery and placement of another BCPS along with ligation of the systemic to pulmonary artery shunt. One month after this, partitioning of the pulmonary arteries was performed to create a classic Glenn shunt, along with placement of a permanent epicardial VVI pacemaker.

Three other patients underwent reoperation 1 to 23 days postoperatively to remove an extra source of pulmonary blood flow, including ligation of systemic to pulmonary artery shunts placed at the time of BCPS ($n = 1$) or at a previous palliative procedure ($n = 1$) and main pulmonary artery ligation ($n = 1$). Two patients underwent median sternotomy debridement 20 and 21 days after BCPS, and another patient underwent re-exploration with diaphragmatic plication 11 days after BCPS. Two patients underwent catheter coil embolization of venous collateral channels 17 and 47 days postoperatively.

Among the variables listed in the Appendix, age <1 month was the only variable significantly associated with early death ($p = 0.03$) and BCPS failure ($p = 0.01$).

At the time of extubation, arterial oxygen saturation was significantly higher than at preoperative catheterization based on the paired *t* test ($p = 0.002$), and did not differ significantly between patients with (83.4 ± 5.7%) and without (81.9 ± 5.0%) accessory pulmonary blood flow. In the operating room after the procedure, the difference in arterial saturations between patients with (85.5 ± 7.9%) and without (78.7 ± 17.3%) accessory pulmonary blood flow had approached significance ($p = 0.11$). This difference narrowed by the time of extubation and is difficult to analyze because patients were receiving different levels of oxygen. There was no significant preoperative to postoperative change in pulmonary artery pressure, but patients with accessory pulmonary blood flow did have significantly higher pulmonary artery pressure than patients without accessory pulmonary blood flow (14.4 ± 3.7 vs. 11.7 ± 2.7 mm Hg, $p = 0.008$). Mean ventricular filling pressure was significantly lower after BCPS (5.3 ± 2.4 mm Hg) than it had been before the operation (7.2 ± 2.3 mm Hg, $p = 0.001$). After BCPS, patients required ventilatory support ranging from 3 to 50 days (median 33 h). The median intensive care unit stay was 4 days (range 1 to 100), and the median postoperative hospital stay was 7 days (range 2 to 109). Patients with accessory pulmonary blood flow required longer ventilatory support (median 60.5 vs. 23.5 h, $p = 0.14$), had a higher incidence of effusions requiring chest tube drainage for longer than 7 days (22% vs. 4%, $p = 0.07$) and were in the intensive care unit (median 5.5 vs. 4.0 days, $p = 0.16$) and hospital (median 10 vs. 7 days postoperatively, $p = 0.21$) longer than patients without additional pulmonary blood flow, although these differences were not statistically significant.

**Late results.** Patients were followed up for a mean of 14.3 ± 11.3 months from BCPS (range 1 to 41). During the follow-up period, four patients underwent a successful extracardiac conduit Fontan procedure 15, 20, 20 and 34 months after BCPS. One patient who underwent BCPS for pulmonary atresia with a ventricular septal defect and a hypoplastic tricuspid valve underwent "one and a half ventricle repair" 20 months after BCPS with conduit reconstruction of the right ventricular outflow tract and tricuspid valve repair.

During the follow-up period there were three late deaths. One patient with polysplenia syndrome died 5 months after BCPS with severe desaturations secondary to multiple pulmonary arteriovenous fistulae (13). A patient with asplenia syndrome died 9 months after BCPS with an undiagnosed respiratory infection. The third patient with hypoplastic left heart syndrome, who underwent tricuspid valvuloplasty 4 months after BCPS and

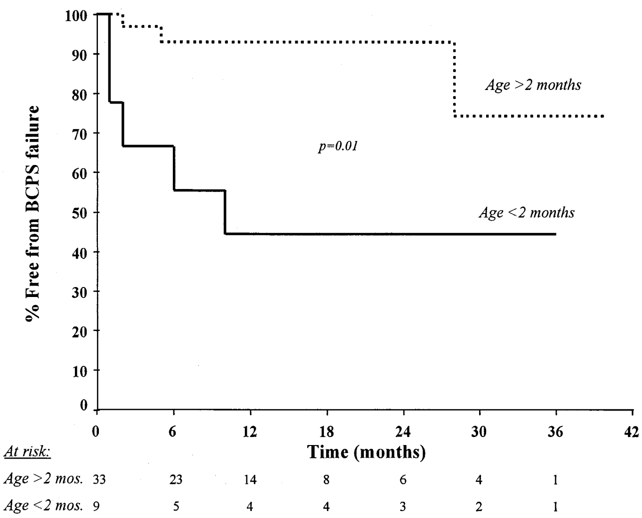


**Figure 2.** Actuarial freedom from bidirectional cavopulmonary shunt (BCPS) failure (death or take-down) for patients <2 and >2 months old at BCPS.

pacemaker placement 5 months after BCPS, died at 6 months because of myocardial infarction (possibly embolic).

Actuarial survival rates were the same at 1 and 2 years, and were 90% for hospital survivors and 86% for all patients. Among patients undergoing primary BCPS, actuarial survival rates at 1 and 2 years were 87% for hospital survivors and 81% for all patients undergoing primary BCPS. Among hospital survivors, there were no significant predictors of poor survival by Cox regression. When configured to incorporate all early and late events (death or take-down), Cox regression analysis found age <2 months ($p = 0.01$, Fig. 2) and pre-BCPS atrioventricular valve regurgitation that was more than mild ($p = 0.016$) to be significantly associated with poorer survival.

At follow-up pulse oximetry or catheterization, mean arterial oxygen saturation was significantly higher than it had been before BCPS ($p = 0.03$). Seventeen patients had undergone follow-up cardiac catheterization. Two of these patients were noted to have pulmonary arteriovenous fistuale (21 and 5 months after BCPS), including one who died from multiple pulmonary arteriovenous fistulae shortly after catheterization and one who underwent Fontan completion.

Follow-up echocardiography was performed in 30 patients, a median of 10 months postoperatively (range 1 to 31), by an echocardiographer who had no knowledge of patient outcomes or the present study. Among the 10 patients with mild preoperative atrioventricular valve regurgitation, four showed improvement—to trace regurgitation in one patient and no regurgitation in three patients. Atrioventricular valve regurgitation remained mild in the other six patients. Of the five patients with moderate atrioventricular valve regurgitation before BCPS, one died early, one continued to have moderate regurgitation and underwent tricuspid valvuloplasty 4 months after BCPS (the patient with hypoplastic left heart syndrome described earlier) and three had improved to mild regurgitation.

## Discussion

Despite the fact that BCPS has been documented to be an effective palliative procedure for patients with most forms of a functional single-ventricle heart (1–7), there have been few reports of its application in young infants (8,9). Concerns about the performance of BCPS in young infants may have to do with the history of unfavorable results after performing a classic Glenn shunt in infants (10–12), along with uncertainty regarding the reactivity of the pulmonary vasculature in this group of patients. Between 1990 and 1995, over one-third (36%) of all BCPS procedures performed at the University of California at San Francisco were in infants <6 months of age. Early results in this cohort of young infants have been similar to those in all patients undergoing BCPS during the same period, with mortality rates of 4.8% and 5.1% and BCPS failure rates of 11.9% and 8.5%, respectively. The only independent risk factor for early death and BCPS failure was age <1 month; and the strongest risk factor for early or late BCPS failure (death or take-down) was age <2 months (Fig. 2). Elevated pulmonary vascular resistance did not apear to be a problem among any of the neonates and very young infants who fared badly after the operation, because they had normal pulmonary artery pressures and transpulmonary gradients throughout their postoperative courses. However, it has been pointed out that pulmonary artery pressures and gradients are not necessarily reliable indicators of resistance when pulmonary blood flow is low or when no good estimate of flow is available. Despite this, persistent desaturation and pulmonary complications were common in this group. Therefore, BCPS does not appear to be a viable option in neonates and should preferably be postponed beyond 2 months of age.

**Role of accessory pulmonary blood flow in BCPS.** A number of recent reports have emphasized the advantages and drawbacks of accessory pulmonary blood flow with BCPS. It has been suggested that accessory pulmonary blood flow may be associated with increased morbidity and mortality after BCPS (9,14,15), whereas post-BCPS arterial oxygen saturation has been found to be slightly higher with accessory pulmonary blood flow (16). In the present study of BCPS performed in infants <6 months of age, accessory pulmonary blood flow was included in 43% of patients, which is lower than in the overall cohort of patients who underwent BCPS during the same period (58%). In the present group of patients, there were no significant differences between patients with and without accessory pulmonary blood flow in terms of survival, BCPS failure, complications or arterial oxygen saturation, either in the immediate postoperative period or at follow-up. Patients with accessory pulmonary blood flow were found to have significantly higher early postoperative pulmonary artery pressures, and analysis revealed trends toward a higher incidence of prolonged effusions (chest tube drainage ≥7 days), longer ventilatory support and longer postoperative intensive care unit and hospital stays in these patients. Thus, short- and intermediate-term analysis reveals no clear advantages to either maintaining or eliminating accessory pulmonary blood flow. Currently, we tend to preserve anterograde pulmonary blood flow in patients with a patent pulmonary outflow tract by banding the main pulmonary artery at the time of construction of the BCPS or by tightening the band if the pulmonary artery has already been banded. In patients with a previous systemic to pulmonary artery shunt, the shunt is generally removed, because the BCPS is performed at the same site. An additional source of pulmonary blood flow (another shunt) is added only if intraoperative arterial blood gases show a partial pressure of oxygen of less than ~30 mm Hg. Among the entire cohort of patients undergoing BCPS during the period of the present study, 58% had accessory pulmonary blood flow, which is higher than the 43% of patients <6 months of age. The primary reason for this is that we add or preserve accessory pulmonary blood flow in almost all patients >3 years of age. Earlier in our experience, we included accessory pulmonary blood flow in a higher percentage of young infants. However, since our experience has shown that pulmonary artery growth and postoperative hemodynamic variables are not compromised in patients without extrapulmonary flow, we have become more conservative in this respect. Nevertheless, the role of extrapulmonary blood flow and the potential complications and benefits of extra flow require further study.

**Role of early BCPS in the staged management of functional univentricular heart.** Our current approach to the management of patients with a functional single-ventricle heart is to perform BCPS within the first 6 months of life, either as the primary procedure in asymptomatic infants after 2 months of age (9) or at 3 to 6 months of age, after placement of a pulmonary artery band or systemic to pulmonary artery shunt, in symptomatic neonates. This minimizes pulmonary overcirculation and volume loading on the single ventricle. We have not identified any pre-BCPS hemodynamic factors that strongly predict for BCPS failure in this cohort of young patients. Patients in this group with pulmonary artery pressures as high as 49 mm Hg and indexed pulmonary vascular resistance as high as 5.9 Wood units underwent successful BCPS, and are doing well with favorable hemodynamic data at follow-up. We do not have strict selection criteria for BCPS in young patients, although we prefer to have pulmonary resistances <2.5 Wood units and a reactive pulmonary vascular bed if resistance is higher than this level. After BCPS, we perform the extracardiac conduit Fontan procedure when the child has grown to the weight of ~15 kg, at which point an adult-sized conduit (20 to 22 mm) can generally be used for the extracardiac inferior cavopulmonary anastomosis. The ages at Fontan completion of the four patients in the present study who underwent the extracardiac Fontan procedure were 20, 24, 25 and 36 months, and all have done well.

**Advantages of early BCPS.** It is generally believed that relieving the volume load on a functional single-ventricle heart by replacing a systemic to pulmonary artery shunt with a BCPS tends to improve atrioventricular valve function in patients with valve insufficiency. In the present study, follow-up analysis showed that atrioventricular valve function had improved in 50% of the 14 patients with mild or moderate pre-BCPS atrioventricular valve regurgitation. This finding, along with recent experimental evidence suggesting that oscillatory work is increased in patients with aortopulmonary shunts (17), adds further support to the argument in favor of converting patients from a systemic to pulmonary artery shunt to BCPS early in life.

**Conclusions.** The BCPS can be performed on an elective basis in infants <6 months of age as a primary or secondary palliative procedure, with results similar to those achieved in older patients. This shunt in patients <6 months of age provides early relief from the ventricular volume loading and elevated pulmonary artery pressures of systemic to pulmonary artery shunting, with no increase in morbidity or mortality, and thus is likely to be a preferable strategy for the early palliation of single-ventricle heart patients. However, BCPS is not recommended in neonates, and in our opinion not in infants <2 months of age. In the <6-month age group, no significant difference in outcomes or functional status was observed between patients with and without accessory pulmonary blood flow.

# Appendix

**Table A1.** Variables Analyzed for Correlation With Early and Late Outcomes

| |
|---|
| Demographic and morphologic variables* |
| Age at operation |
| Continuous |
| <1 mo (yes/no) |
| <2 mo (yes/no) |
| <3 mo (yes/no) |
| Weight at operation |
| Previous palliative procedure (yes/no) |
| Previous systemic to pulmonary artery shunt (yes/no) |
| Diagnosis |
| Hypoplastic left heart syndrome (yes/no) |
| Heterotaxy single ventricle (yes/no) |
| Catheterization and angiographic variables (before BCPS) |
| Arterial oxygen saturation |
| Mean pulmonary artery blood pressure |
| Ventricular (dominant/single) end-diastolic pressure |
| Indexed pulmonary blood flow |
| Pulmonary to systemic blood flow ratio |
| Indexed pulmonary vascular resistance |
| Pulmonary artery index (indexed to body surface area) |
| Total (bifurcation, lower lobe) |
| Right (bifurcation, lower lobe) |
| Left (bifurcation, lower lobe) |
| Atrioventricular valve regurgitation |
| Mild or greater |
| Moderate or greater |
| Operative variables |
| Bilateral BCPS (yes/no) |
| Pulmonary artery augmentation (yes/no) |
| Intracardiac procedures performed (yes/no) |
| Accessory pulmonary blood flow (yes/no) |
| Cardiopulmonary bypass (yes/no) |
| Time |

*Also analyzed for correlation with preoperative catheterization and angiographic variables. BCPS = bidirectional cavopulmonary shunt.

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