Pulmonary artery stenosis in hybrid single-ventricle palliation: High incidence of left pulmonary artery intervention

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Objective: Pulmonary artery growth is an important determinant of outcome in single-ventricle strategies. Higher rates of pulmonary artery intervention have been reported with hybrid-based palliation when compared with Norwood palliation.

Methods: We performed a retrospective review of pulmonary artery growth and clinical outcomes in patients undergoing hybrid-based single-ventricle palliation.

Results: The stage I hybrid procedure was performed in 72 patients between 2004 and 2012, of whom 54 were on a Fontan palliative pathway. Thirty-four infants completed stage II, and 20 infants underwent the Fontan operation. The mean diameters of the right pulmonary artery (5.6 ± 1.9 mm) and left pulmonary artery (5.6 ± 2.1 mm) were similar before stage II. After stage II, the right and left pulmonary artery diameters were 8.5 ± 2.1 mm and 5.8 ± 1.3 mm, respectively ($P < .001$), and after the Fontan operation, these were 8.8 ± 2.0 mm and 6.4 ± 1.1 mm, respectively ($P = .002$). The mean right pulmonary artery z score was normal throughout, but the left pulmonary artery did not maintain a normal size. The cumulative pulmonary artery intervention rate was 50% at any time after stage II. Fifteen interventions (88%) were performed after stage II (35% during the same hospitalization, 71% <60 days). The most intervened site was the midsection of the left pulmonary artery (41%). Initial pulmonary artery intervention was balloon dilation in 59% of patients and stent implantation in 41% of patients. Half of patients with initial balloon dilation required reintervention.

Conclusions: There is significant risk of left pulmonary artery compromise after the second stage of hybrid palliation associated with a high intervention rate. (J Thorac Cardiovasc Surg 2015;149:1102-10)

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See related commentary on page 1111.

Supplemental material is available online.

The hybrid approach (bilateral pulmonary artery [PA] banding, ductal stenting) for single-ventricle hearts is an alternative to the Norwood procedures (right ventricle [RV]-PA conduit and modified Blalock–Taussig shunt [BTS]) with comparable midterm results.[1-6] Its theoretic advantage is avoidance of cardiopulmonary bypass surgery, hypothermic circulatory arrest, and the systemic inflammatory reaction during the neonatal period, which may have long-term implications, particularly for the heart, lungs, and brain.

The complication of PA obstruction has negative clinical implications on the course to and after the Fontan operation.[1,5-13] Recent studies report smaller PA sizes and a higher incidence of PA intervention in those undergoing the hybrid approach compared with the Norwood procedure after the second operation.[5,6,14] The PA banding and de-banding process is unique to the hybrid procedure. Excessive banding could impede blood flow and vessel growth. Band migration may compromise lobar branches. During the second stage of hybrid surgery (Damus–Kaye–Stansel anastomosis and bidirectional cavopulmonary connection [BCPC]), the PA band may necessitate more extensive reconstruction of the superior vena cava (SVC) to PA anastomosis and branch PAs. Band-induced alterations in the vessel wall architecture may predispose to obstruction.[15,16] There may be space constraints for the PA from the newly constructed aorta and adjacent bronchus, potentially further compounded by the rigidity of any ductal stent remnant[12,13] (Figure 1).

The objective of this study was to perform a detailed review of the PAs of patients who underwent the hybrid procedure for single-ventricle palliation with respect to longitudinal growth, incidence, and timing of PA obstruction, methods of intervention, and impact on clinical outcome.

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Abbreviations and Acronyms

BCPC = bidirectional cavopulmonary connection
BTS = Blalock–Taussig shunt
HLHS = hypoplastic left heart syndrome
LLI = lower lobe index
LPA = left pulmonary artery
PA = pulmonary artery
RPA = right pulmonary artery
RV = right ventricle
SVC = superior vena cava

MATERIALS AND METHODS

Patient Population

This retrospective study was approved by the hospital research ethics board. Patient selection for the hybrid palliation was decided on a case-by-case basis at our interdisciplinary conference. Between January 2004 and December 2012, a stage I hybrid procedure was performed in 72 patients. No patient underwent a hybrid procedure as a bridge to a Norwood operation. Patients in whom the hybrid stage I procedure was performed as a bridge to transplantation or biventricular repair or as a "salvage" procedure were excluded because serial PA data were not available. A salvage procedure was performed for an infant with prohibitive risks for any type of first-stage single-ventricle palliation. Subanalyses were performed for selected anatomic features, specifically hypoplastic left heart syndrome (HLHS), bilateral SVCs, and right aortic arch.

Our institution has previously described the surgical and interventional techniques used for hybrid single-ventricle palliation.[3,13,17] Inter-stages I and II are defined as the period between hybrid stages I and II and between stage II and Fontan operations, respectively. Hospital records, echocardiographic and magnetic resonance imaging, cardiac catheterization, and surgical records were reviewed.

Evaluation of Pulmonary Arteries

Angiography was routinely performed during the stage I hybrid procedure, before stage II and Fontan operations, and after Fontan completion. Starting in September 2008, exit angiography was routinely performed during stage II surgery. These and any additional angiograms were reviewed. The central left pulmonary artery (LPA) and right pulmonary artery (RPA) were measured at 3 points: immediately proximal to the first branch and at the narrowest and largest diameters. The PA diameter reported in the "Results" section refers to the site proximal to the first lobar branch. PA diameters are expressed as z scores.[18] Range in central PA size was determined by subtracting the smallest from the largest diameter. Nakata index was calculated.[19] PA stenosis was defined as greater than 50% narrowing of the blood vessel diameter compared with the adjacent vessel proximally or distally. Diameters of the lower and upper lobe branches were measured. Lower lobe index (LLI) was calculated as the sum of the right and left lower lobe branch cross-sectional areas indexed to body surface area. Symmetry of PAs was evaluated by the ratios of the RPA and LPA diameters and RPA and LPA LLIs. The distance between the PA band and the origin of the first lobar branch was measured at the initial hybrid procedure and the pre-stage II catheterization to determine interval band migration. Migration was defined as being significant if the band caused stenosis or occlusion of a lobar branch. To test our hypothesis of LPA external compression by the reconstructed aorta, aortic measurements were made at the levels of the ascending aorta (neo-aorta), transverse arch (distal to innominate artery), distal arch (at the base of the left common carotid artery), and the diaphragm. A single investigator (OR) obtained all measurements.

Norwood Cases

During the same period, 88 patients underwent a Norwood stage I procedure (BTS, n = 77; Sano shunt, n = 11). PA intervention rates for these patients are reported.

Statistical Analysis

Data are presented as mean ± standard deviation unless otherwise stated. PA intervention and clinical outcome were evaluated with respect to the variables of PA size (absolute and relative), neo-aorta dimensions, PA indices, and LPA/neo-aorta diameter ratio, the narrowest LPA diameter compared with the neo-aorta at all stages. Patients with right aortic arch were excluded from LPA/neo-aorta analysis. Change in PA diameter or "growth" of the branch PAs was analyzed using a paired Student t test. The differential growth of the RPA and LPA after the initial hybrid

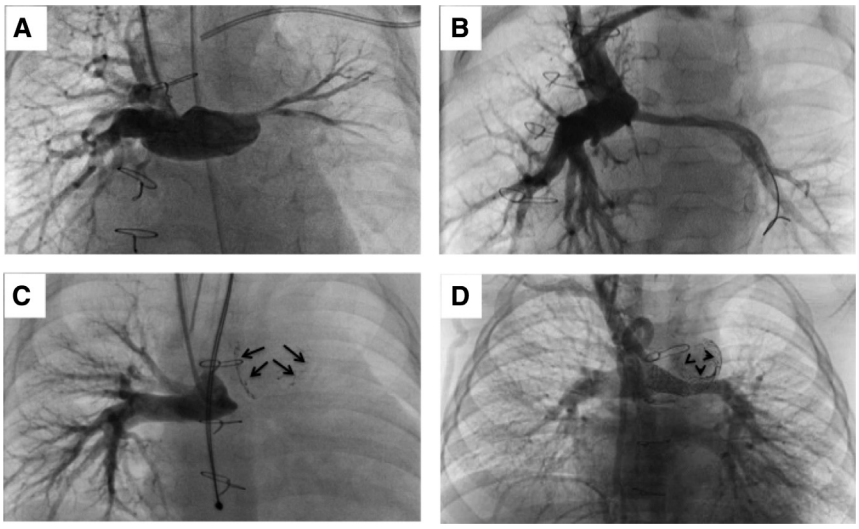

FIGURE 1. Angiograms representative of PA problems after stage II hybrid surgery are shown. A, Severe stenosis distal to LPA patch. B, Asymmetric PA tree. Note the diffusely hypoplastic LPA and well-developed RPA. C, Total occlusion of LPA (*arrows*; retained ductal stent). D, Proximal and mid-section stenosis of LPA with previously implanted LPA stent. Note the retained ductal stent (*arrowheads*) at the level of LPA obstruction.