Evaluation of bilateral pulmonary artery banding for initial palliation in single-ventricle neonates and infants: risk factors for mortality before the bidirectional Glenn procedure

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Abstract

OBJECTIVES: Bilateral pulmonary artery banding is considered as ‘first-stage’ palliation for neonates who have hypoplastic left heart syndrome. This study aimed to identify risk factors that influence outcome before the bidirectional Glenn operation.

METHODS: This retrospective evaluation involved 30 consecutive patients with hypoplastic left heart syndrome, or a variant, who underwent bilateral pulmonary artery banding between August 2005 and December 2011 at our institution. Clinical echocardiographic, operative and catheter examination data were reviewed.

RESULTS: This study included 9 patients with hypoplastic left heart syndrome and 21 patients with variants. Bilateral pulmonary artery banding was performed at a median age of 7 days. Finally, 19 patients had the bidirectional Glenn operation performed (Group A), and the remaining 11 patients died before the bidirectional Glenn procedure (Group NA). Catheter evaluations before the bidirectional Glenn procedure were carried out at 97 ± 34 days. The mean pulmonary venous wedge pressure was significantly lower (Group A: 13.1 ± 3.1 mmHg vs Group NA: 22.9 ± 3.7 mmHg, P < 0.01), systemic ventricular ejection fraction was higher (54.4 ± 10.7 vs 41.7 ± 9.9%, P < 0.05), systemic ventricular end-diastolic pressure was lower (6.1 ± 2.4 vs 10.5 ± 3.6 mmHg, P < 0.05) and the rate of patients with more than mild systemic atrioventricular valve regurgitation was lower in Group A than in Group NA (15.7% vs 62.5%, P < 0.05). Multivariate logistic regression analysis showed that mean pulmonary venous wedge pressure was the most significant predictor of attaining the bidirectional Glenn anastomosis (odds ratio: 2.35, P < 0.01).

CONCLUSIONS: Postoperative atrioventricular valve regurgitation, cardiac function and mean pulmonary venous wedge pressure are closely correlated with mortality after bilateral pulmonary artery banding. Additional treatments, including operations, are considered to maintain cardiac function and not to raise pulmonary venous wedge pressure before the bidirectional Glenn procedure.

Keywords: Congenital heart disease • Hypoplastic left heart syndrome • Risk factor • Bilateral pulmonary artery banding • Bidirectional Glenn procedure

INTRODUCTION

Patients with hypoplastic left heart syndrome (HLHS), including variants of HLHS and other complex congenital cardiac defects with functional single ventricle physiology and systemic outflow tract obstruction, continue to be a challenge to even the most experienced congenital heart surgeons [1, 2]. Although operative mortality has steadily decreased from when Norwood et al. performed and reported their first successful procedure in 1981 [3], high-risk neonates undergoing first-stage Norwood palliation still face mortality rates of 20–50% [4–6]. According to published data, low birth weight, prematurity, preoperative shock, renal failure, small ascending aorta (<0.3 cm), restrictive atrial communication, extracardiac abnormalities and chromosomal abnormalities are risk factors for mortality after the Norwood operation [2, 4, 6–9]. Improved understanding of the postoperative physiology, and experience with surgical techniques and perioperative care of patients with HLHS have resulted in improved outcomes [10].

Bilateral pulmonary artery banding (bPAB) has recently attracted attention because it is less invasive than surgery, requiring cardio-pulmonary bypass support [9, 11, 12]. Ductal stenting or continuance of prostaglandin E1 are strategies that have been used for the initial palliation of HLHS until second-stage palliation is undertaken [13, 14].

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We have performed bPAB as the first-stage palliation in HLHS patients, or in those with its variants, followed by the Norwood procedure with a systemic-to-pulmonary arterial shunt or Glenn anastomosis, since 2005. However, some patients were not able to attain bidirectional Glenn (BDG) anastomosis. In the present study, we evaluated the outcome of these procedures to clarify the risk factors for not achieving Glenn anastomosis.

PATIENTS AND METHODS

Patients

All of the patients or their families gave their written informed consent. We performed a retrospective review of 30 patients with anatomical or functional single ventricle anatomy who underwent bPAB and ductal stents or maintenance of prostaglandin E1 from August 2005 to December 2011 at our institution. All clinical, echocardiographic, angiographic and operative data were reviewed. We defined Group A as patients who had a successful BDG operation with or after the Norwood procedure. Group NA included patients who died before the BDG operation. The patients’ characteristics, such as age at the bPAB procedure, circumference of the bPAB tape, systolic blood pressure, oxygen saturation after bPAB and systemic atrioventricular regurgitation, and data of catheter examinations before the BDG procedure were compared between survivors (Group A) and non-survivors (Group NA). Risk factors for hospital death with failure to achieve Glenn anastomosis were assessed. Follow-up was complete for all 30 patients (100%).

Bilateral pulmonary artery banding technique

bPAB was performed in the operating room with patients under general anaesthesia. After standard median sternotomy and minimal mobilization of pulmonary artery branches, tape for bPAB was made comprising a 0.4-mm polytetrafluoroethylene patch. The patch was cut longitudinally to create a strip 1–2 mm wide for all cases. The right and left pulmonary arteries were individually banded to ~70 and 80%, respectively, of the normal pulmonary artery branch diameter. The velocity of the pulmonary artery banding position was measured by echocardiography in the operative field, which was adjusted to ~3.0 m/s. Right atrial lines, pacing wires and pericardial drain tubes were placed. The sternum was closed unless the patient had considerable intraoperative dysrhythmia or anasarca.

Echocardiographic and catheter examinations

Echocardiography was performed in the intensive care unit or cardiac ward. The degree of regurgitation was graded on an ordinal scale from 0 to 4 based on available transthoracic echocardiographic reports (0 = no regurgitation, 1 = trace or trivial, 2 = mild, 3 = moderate and 4 = severe). The degree of regurgitation was estimated before the bPAB operation, after the bPAB operation and before the BDG procedure. Cardiac catheterizations were performed after the bPAB operation and just before the BDG procedure. When cardiac catheterizations were performed several times, data from the most recent study were used.

Statistical analysis

Data are shown as numbers and percentages, or the mean ± standard deviation. Dichotomous variables were analysed with Fisher’s exact test, and continuous variables were analysed with Student’s t-test. Univariate analysis was used to identify significant variables, which were then entered in a stepwise fashion into multivariate analyses. Data were analysed using JMP 9.0 (SAS Institute, Cary, NC, USA). Any P value less than 0.05 was considered to have statistical significance.

RESULTS

Patient population

Between August 2005 and December 2011, 30 patients (17 boys and 13 girls) received the bPAB procedure for HLHS and HLHS variants as first palliation by two surgeons. Nine patients had HLHS. The remaining 21 had variants of HLHS with left ventricular, aortic valvular or aortic arch hypoplasia, with the following diagnosis: ventricular septal defect with coarctation of the aorta or interrupted aortic arch (n = 9), double outlet right ventricle with critical aortic stenosis, coarctation of the aorta or interrupted aortic arch (n = 4), heterotaxy syndrome with coarctation of the aorta (n = 6), unbalanced atrioventricular septal defect (n = 1), and tricuspid atresia with transposition of the great arteries and coarctation of the aorta (n = 1). Among the 9 patients with HLHS, 5 (55.6%) had aortic stenosis and mitral stenosis, 3 (33.3%) had aortic atresia and mitral atresia, and 1 (11.1%) had aortic atresia and mitral stenosis. Among them, aortic atresia was present in 4 patients (44.4%). The median diameter of the ascending aorta was 2.8 mm (range, 1.8–3.5 mm).

Cardiac and extracardiac complications, and chromosomal abnormalities are shown in Table 1. The median weight of the patients at the bPAB operation was 2657 g (range, 1018–3616 g). Twelve patients (40%) weighed less than 2.5 kg at birth, including 4 patients (13%) who weighed less than 1.5 kg. Seven patients (23%) were premature (gestational age, <37 weeks). Sex, gestational age, prematurity, body weight at birth, low birth weight, prenatal diagnosis, preoperative shock and isomerism were not associated with mortality before the BDG procedure. Only the presence of complications (shown in Table 1) was significantly related to achievement of the BDG procedure after bPAB (P <0.05, Table 2).

Bilateral pulmonary artery banding operation

The median age at the bPAB operation was 7 days old (range, 0–108 days: ≤7 days old, n = 17; ≤14 days old, n = 6; ≤30 days old, n = 3; >30 days old, n = 2). Five patients (17%) were older than 14 days old at the time of their initial palliation. None of the parameters concerning the bPAB operation were associated with mortality before the BDG procedure (Table 3). None of the patients had intraoperative arrhythmia or anasarca, precluding sternal closure. After our bPAB procedure, the mean oxygen saturation of arterial blood as measured by an oxymeter was 83.8% (65–97%) under the condition of 21–60% oxygen concentrations in the operative room.
Follow-up

After bPAB, all of the 30 patients were followed for a mean of 40.2 months (median, 42 months; range, 6–98 months). Among these 30 patients, 19 (63%) survived the BDG procedure (Fig. 1). Among them, 18 patients underwent BDG with the Norwood procedure, and 1 patient underwent the BDG procedure 355 days after the Norwood procedure with a right ventricle to pulmonary artery (RV-PA) shunt. The median age to the BDG procedure was 148.7 days after birth (range, 89–703 days). Ten patients underwent the Fontan procedure and 7 patients are waiting for the Fontan procedure. Two patients died from heart failure during the inter-stage period between the BDG and Fontan procedures.

There were 9 hospital deaths (30%) after bPAB. Among these 9 patients, the cause of death included cardiac heart failure (n = 4), sudden death after bPAB (n = 2), pulmonary hypertension (n = 1), necrotizing enterocolitis (n = 1) and coronary ischaemia (n = 1).

There were two inter-stage deaths after the Norwood procedure with an RV-PA shunt resulting from cardiac failure (n = 2).

Echocardiographic and catheter examinations

There were three early deaths before postoperative catheterization. Cardiac catheterizations before the BDG procedure were performed at a mean (standard deviation, SD) of 97 ± 34 days. By univariate analysis, the predictor for mortality before the BDG procedure was systemic atrioventricular regurgitation of degree 2 or greater (Group A: 15.7% vs Group NA: 62.5%, P < 0.05). The morphology of systemic atrioventricular valves included 7 mitral valves, 16 tricuspid valves and 4 common atrioventricular valves. Before the BDG procedure, 9 patients had Grade 0 atrioventricular valve regurgitation, 10 had Grade 1, 4 had Grade 2, 3 had Grade 3 and 1 had Grade 4 in echocardiographic examinations.

In catheter examinations, predictors for mortality before the BDG procedure were mean pulmonary venous wedge pressure (Group A: 13.1 ± 3.1 mmHg vs Group NA: 22.9 ± 3.7 mmHg, P < 0.01), systemic ventricular ejection fraction (Group A: 54.4 ± 10.7% vs Group NA: 41.7 ± 9.9%, P < 0.05) and systemic ventricular end-diastolic pressure (Group A: 6.1 ± 2.4 mmHg vs Group NA: 10.5 ± 3.6 mmHg, P < 0.001).
P < 0.05). Mean pulmonary venous wedge pressure was an independent predictor of overall mortality by multivariate logistic regression analysis (P = 0.003; odds ratio, 2.35) (Table 4).

DISCUSSION

Despite a significant improvement in mortality associated with Stage 1 Norwood palliation in recent years, this form of surgical palliation for neonates still remains a high-risk operation. Recently, a more refined and collaborative effort to implement a hybrid strategy, comprising bPAB with continuous lipo-prostaglandin E1 administration or ductal stenting, has emerged as an alternative form of palliation for HLHS or associated anomalies [9, 15–17]. In addition, Akintuerk et al. reported a successful combined Stage 2 surgical procedure, which has opened the path from hybrid palliation towards a Fontan circulation, and this approach continues to gain interest [15].

Increased awareness of inter-stage mortality as an important contributor to deterioration in patients, an increased risk of associated comorbidities and the challenging technical aspects of Stage 2 reconstruction require a longitudinal and comprehensive analysis to gauge the overall success of any management strategy in these patients. In our institution, we perform the bPAB procedure as a less invasive first-stage palliation than the Norwood procedure. We then shift to cardiopulmonary bypass later in life at an age when a circulation in series can be established. However, some patients die after bPAB before having the BDG procedure.

Our study investigated risk factors for not achieving Glenn anastomosis. Inter-stage mortality in the bPAB strategy is in the range of 5–20% for patients with a typical and high risk [9, 13]. In our study, the inter-stage mortality rate was 30% (9 patients) and 2 patients died between the Norwood procedure and the BDG procedure. Some patients died because of continuous heart failure, pulmonary hypertension and coronary events, even if we performed bPAB as primary palliation to restrict pulmonary blood flow. These results suggest that we need to identify these cases earlier for interventions according to the parameters after bPAB. The results of our patients indicated that we should maintain lower

Table 4: Echocardiographic and catheterization data before the Glenn procedure

<table>
<thead>
<tr>
<th></th>
<th>Group A</th>
<th>Group NA</th>
<th>Univariate P-value</th>
<th>Multivariate P-value</th>
<th>95% CI</th>
<th>OR</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at catheterization (days)</td>
<td>94.2 ± 28.7</td>
<td>106.1 ± 46.2</td>
<td>0.52</td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>SAVVR (≤mild) on echocardiography</td>
<td>319</td>
<td>5/8</td>
<td>0.015</td>
<td>0.96</td>
<td>0.286–0.958</td>
<td>1.54</td>
</tr>
<tr>
<td>SaO2 (%)</td>
<td>71.9 ± 6.8</td>
<td>76.3 ± 7.7</td>
<td>0.20</td>
<td></td>
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<td></td>
</tr>
<tr>
<td>PV wedge pressure (mmHg)</td>
<td>13.1 ± 3.1</td>
<td>22.9 ± 3.7</td>
<td>0.0005</td>
<td>0.003</td>
<td>0.286–0.958</td>
<td>2.35</td>
</tr>
<tr>
<td>Op/Qs</td>
<td>1.1 ± 0.5</td>
<td>1.4 ± 0.7</td>
<td>0.36</td>
<td></td>
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<tr>
<td>SVEF (%)</td>
<td>54.4 ± 10.7</td>
<td>41.7 ± 9.9</td>
<td>0.015</td>
<td>0.89</td>
<td>0.286–0.958</td>
<td>0.92</td>
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<tr>
<td>PA index (mm²/m²)</td>
<td>6.1 ± 2.4</td>
<td>10.5 ± 3.6</td>
<td>0.011</td>
<td>0.66</td>
<td>0.286–0.958</td>
<td>1.62</td>
</tr>
<tr>
<td></td>
<td>226.6 ± 120.9</td>
<td>328.3 ± 221.2</td>
<td>0.32</td>
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</table>

SAVVR: systemic atrioventricular valve regurgitation; PV: pulmonary vein; SVEF: systemic ventricular ejection fraction; SVEDP: systemic ventricular end-diastolic pressure; PA: pulmonary artery; CI: confidence interval; OR: odds ratio.

Figure 1: Summary of outcomes for all patients. HLHS: hypoplastic left heart syndrome; PA: pulmonary artery; RV-PA: right ventricle to pulmonary artery; TCPC: total cavopulmonary connection.
pulmonary venous wedge pressure, and higher end-diastolic pressure and ejection fraction of the systemic ventricle, with less atrioventricular valve regurgitation after the bPAB procedure (Table 3). We performed additional valve repairs (Kay’s method in 2 patients and De Vega anuloplasty in 2 patients in our institution) and an earlier second-stage operation for patients who could not control their cardiac failure after bPAB.

Some variables have been shown as risk factors for mortality before the BDG procedure. Low body weight, prematurity, preoperative shock, small ascending aorta, significant tricuspid regurgitation and extracardiac abnormalities are poor prognostic factors for survival, as shown previously [18–22]. In our study, prematurity, low birth weight, circumference and the age at the bPAB procedure were not significantly associated with mortality. However, mean pulmonary venous wedge pressure, systemic atrioventricular regurgitation, ejection fraction and end-diastolic pressure were significantly associated with mortality before the BDG procedure. Based on our lack of finding of significant differences in previously reported risk factors for mortality before the BDG procedure (e.g., low birth weight and aortic atresia), we consider it important to preserve cardiac function after bPAB. Therefore, maintaining an open patent ductus arteriosus and blood flow to the coronary artery and brain are necessary, especially for those who have the morphology of aortic atresia. We frequently monitored using echocardiographic examinations whether the patent ductus arteriosus was closed. With regard to low birth weight, we consider that patients who undergo bPAB with low birth weight can attain the BDG anastomosis, similar to those with an appropriate weight, if cardiac function is maintained.

Three patients underwent the Norwood procedure early with pulmonary artery angioplasties and creation of an atrial septal defect before the BDG procedure. One of these patients underwent the BDG procedure 12 months later. The other 2 patients died because of cardiac failure after the Norwood procedure with an RV-PA shunt. In these patients, we should have considered earlier interventions, including atrioventricular valve repair and outflow reconstruction, to control their heart failure after bPAB procedure. These findings were from a small number of cases, but suggest that additional surgical procedures are effective after bPAB.

Study limitations

There are several limitations to this study, which was retrospective and had a small number of patients. Therefore, this study had all of the limitations inherent to such a design. The availability of data was variable. Patients were not managed in a standardized fashion and, therefore, we did not make a decision on the timing of the second operation.

CONCLUSION

Postoperative atrioventricular valve regurgitation, cardiac function and mean pulmonary venous wedge pressure are closely correlated with mortality after bPAB. These findings suggest that earlier interventions, such as a secondary operation, including valve repair and outflow reconstruction, should be considered for patients experiencing circulation management after bPAB. This is important for maintaining cardiac function and not increasing the pulmonary venous wedge pressure before the BDG procedure.

Conflict of interest: none declared.

REFERENCES


