Forward Flow Through the Pulmonary Valve After Bidirectional Cavopulmonary Shunt Benefits Patients at Fontan Operation

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Background. The impact of additional pulmonary forward flow (APF) through the pulmonary valve at the time of bidirectional cavopulmonary shunt (BCPS) is unknown.

Methods. Between 2000 and 2010, 276 patients had BCPS and 126 of them were selected, including 60 patients with APF via pulmonary valve and 66 patients, in whom the pulmonary valve was closed. We compared the length of hospital stay and duration of pleural drainage at BCPS and Fontan operations. We also compared the number of surgical interventions before BCPS, the number of operations between BCPS and Fontan operation, Nakata index prior to Fontan operation, grade of atrioventricular valve regurgitation (AVVR), and oxygen saturations prior to Fontan operation.

Results. Prior to BCPS, 20% (12 of 60) of patients with APF and none without APF had pulmonary artery (PA) banding. More patients without APF had systemic-to-PA

The impact of leaving the additional pulmonary for-**L** ward flow (APF) across the stenosed pulmonary valve at the time of bicaval pulmonary artery shunt (BCPS) remains unknown [1-5]. The BCPS with APF may potentially prolong duration of pleural effusion, increase the ventricular volume overload and atrioventricular valve regurgitation. On the other hand, BCPS with APF may increase oxygen saturations, enhance the growth of pulmonary arteries, and preclude development of arteriovenous fistula in the lung by exposing the lung with the hepatic venous flow. Although previous papers [3, 4] suggested that BCPS with APF is not associated with adverse outcomes, the impact of APF on patients with BCPS undergoing Fontan operation is not clear. This retrospective study was undertaken to assess the impact of the APF in patients operated at a single institution.

shunts (p < 0.01). Fontan operation was completed in 58% (35 of 60) of patients with APF and in 68% (45 of 66) of patients without APF (p = 0.34). There was no difference in the length of hospital stay or duration of pleural drainage at BCPS. No significant difference was observed in the number of surgical procedures between BCPS and Fontan operation, grade of AVVR or oxygen saturations before Fontan operation. Children with APF had a higher Nakata index (p = 0.02) prior to Fontan operation, shorter duration of pleural drainage (p = 0.009) and shorter hospital stay (p = 0.009) after Fontan operation.

Conclusions. Children with APF at BCPS had better developed PAs, shorter duration of pleural drainage, and shorter hospital stay after Fontan operation.

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Patients and Methods

A retrospective study of 276 patients was done to review those who underwent BCPS with or without APF from 2000 to 2010. This study was approved by the Ethics Committee of The Royal Children's Hospital. Patient consent was waived because of the retrospective nature of the study.

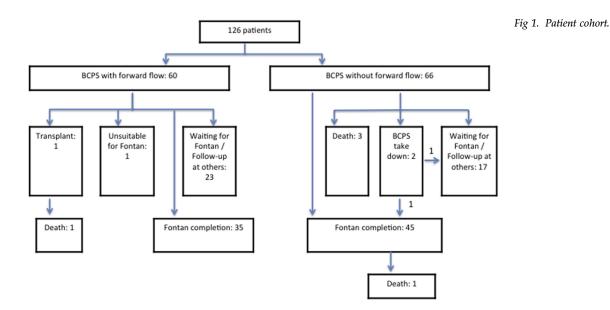
Patients who had a Kawashima operation (n = 5), Damus-Kaye-Stansel anastomosis or Norwood operation (n = 123), or Ebstein anomaly repair (n = 3), one and a half circulation (n = 5), and others (n = 12) were excluded. Two additional patients were excluded because they were adults at the time of BCPS; aged 32 and 37 years. Thus, 126 children were selected in the study, including 60 children with APF and 66 children without APF at the time of BCPS (Fig 1). An extracardiac Fontan operation was performed as previously described [6]; the main

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pulmonary artery (PA) was closed at the time of Fontan if it remained.

Statistics

Patient characteristics were summarized using mean, standard deviation, median, minimum, and maximum for continuous variables, and counts and percentages for categoric variables. The Fisher exact test was used to compare categoric outcomes between the BCPS with forward flow and BCPS without forward flow groups. Due to the non-normal distribution of most of the continuous outcomes, the log and log-log transformations were used to normalize the distribution in order to apply the *t* test. The Wilcoxon rank sum test was also used to compare medians on non-transformed data to obtain mean estimates of the difference between the 2 groups in Nakata (PA) index prior to the Fontan operation. The duration of drainage and length of hospital stay after Fontan operation were also compared. Generalized estimating equation (GEE) models were applied due to the non-normal distribution of those variables. The GEE models are similar to regression models and are usually adopted for longitudinal and dependent data and do not assume normality of the data for continuous outcomes. The GEE can also be applied to non-longitudinal data and allow for covariate adjustment in the estimates of mean difference between the 2 groups. Overall survival was estimated using the Kaplan-Meier method and was measured from the date of BCPS to death due to any cause. Survival for subjects who were still alive at the date of last follow-up was censored. The logrank test was used to compare overall survival between the 2 groups. All analyses were performed in R version 3.1.2 (R Foundation for Statistical Computing, Vienna, Austria).

Results

Median age at BCPS was 1 year (range, 0.14 to 14.7) in patients with APF and 0.85 year (range, 0.14 to 14.7) in

patients without APF (p = 0.33). Between the 2 groups there were differences in the distribution of diagnoses: pulmonary atresia and intact ventricular septum with 0% (0 of 60) in patients with APF versus 26% (17 of 66) in patients without APF (p < 0.001) and pulmonary stenosis with 35% (21 of 60) in patients with APF versus 18% (12 of 66) in patients without APF (p = 0.042) (Table 1). Median number of surgeries prior to BCPS was lower in patients with APF (Table 2): 0, 0 in 52% (31 of 60); 1 in 27% (16 of 60); 2 in 20% (12 of 60); 3 in 2% (1 of 60), as compared to patients without APF: 0 in 11% (7 of 66); 1 in 55% (36 of 66); 2 in 29% (19 of 66); and 3 in 6% (4 of 66) (p < 0.001). Before BCPS, children without APF more often had Blalock-Taussig shunts (BTS) (64% [42 of 66] vs 20% [12 of 60], *p* < 0.001) and central shunts (23% [15 of 66] vs 5% [3 of 60], p = 0.005). Some children (13%, 9 of 66) in a group of BCPS without APF did not have any palliative shunts prior to the BCPS and had the main pulmonary artery divided at the time of BCPS. The BCPS with APF had more pulmonary artery banding (20% [12 of 60] vs 0% [0 of 66], *p* < 0.001).

Median age at Fontan (5.4 years [range, 3.8 to 16.7] vs 5.3 years [range, 3.1 to 14.0], p=0.33), interval period from BCPS to Fontan (4.1 years [range, 0 to 11.8] vs 4.4 years [range, 1.9 to 12.1], p = 0.95), and Fontan completion rate (58% [35 of 60] vs 68% [45 of 66], p = 0.34) were not different between the 2 groups.

Mortality and Morbidity From BCPS to Fontan Operation

There was no significant difference between the 2 groups in freedom from death (Table 3, Fig 2). Among patients in the with APF group, there was 1 death. The patient, who had a diagnosis of double outlet right ventricle, had received an orthotopic heart transplant 3 years after the BCPS and died 5 years after the BCPS. Three children died in the children without APF group. Two patients died 7 days and 8 days after the BCPS

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