In Hospital Outcomes after Bidirectional Cavopulmonary Anastomosis: 18 Year Experience from a Single Center.

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Introduction

Bidirectional cavopulmonary shunt (BCPS) is used for the interim palliation of a variety of cyanotic cardiac lesions.1 It is a standard palliative step for patients with functionally univentricular hearts in hope of achieving Fontan completion. At our center, first case of BCPS was performed on February 7, 2002. Here, we share our experience with BCPS over last 18 years.

Methods: This is a retrospective analysis of all patients undergoing BCPS from February 2002 to July 2019. Patients who underwent BCPS as a part of one-and-half ventricular repair for Ebstein’s anomaly were excluded. Baseline, intraoperative, and postoperative variables were collected from hospital records.

Results: A total of 326 patients with mean age of 5.7±5.8 years (median 3.5 years; range: 1.5 months-32 years) underwent BCPS over last 18 years. Majority (61%) were males. Double outlet right ventricle (DORV) was the most common primary cardiac lesion (30%) followed by tricuspid atresia (23%). Mean ICU stay was 3.9±4.8 days, with hospital stay of 7.8±5.9 days. In-hospital mortality was 15%. Patients who died had low body weight (11.8±10.7 kg vs 16.0±11.6 kg; p=0.019), and longer cardiopulmonary bypass time (101±64 min vs 76±42 min; p=0.001). Oxygen saturation improved significantly at the time of discharge (79.2±9.5% vs 68.6±13.7%; p=0.000).

Conclusion: This is the first report of its kind to analyze the outcomes of BCPS in our center. We have discussed the evolution of BCPS surgery in our center and presented our outcomes. Our in-hospital mortality remains high, and we need to strive towards reducing the mortality.

Abstract

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Keywords: Bidirectional cavopulmonary shunt; staged palliation; univentricular heart.

DOI: https://doi.org/10.3126/njh.v17i2.32673
being major risk factors of in hospital mortality. We performed our first case of BCPS on February 7, 2002 (2058/10/25 BS); following which we have performed hundreds of this palliative procedure. Here, we share our in hospital outcomes following BCPS over last 18 years.

Methods
This is a retrospective analysis of all patients who had undergone bidirectional cavopulmonary shunt (BCPS) in our center from February 2002 to July 2019. Patients undergoing BCPS as a part of one-and-half ventricular repair for Ebstein’s anomaly were excluded; however, isolated BCPS for Ebstein’s anomaly were included. Operation theatre (OT) record files, anesthesia record files, intensive care unit (ICU) record files, and discharge files were analyzed for relevant information. In case of missing data, the analysis was performed considering the number of available data alone. Statistical analysis was done using SPSS version 26 (IBM SPSS Statistics for Windows, Armonk, NY: IBM Corp). Data were expressed using mean±SD, median, range, and percentage wherever appropriate. Independent sample t test, and paired sample t test were utilized depending on the variables. A p value of <0.05 was considered as significant.

Results
We performed our first case of BCPS on February 7, 2002 (2058/10/25 BS). Following that landmark surgery, 326 patients underwent BCPS in our center over the last 18 years. It took more than six years to reach up to our 50th case; following which we started to see steep rise in the number of cases (Figure 1).

Baseline characteristics of all patients are presented in Table 1. Mean age of the patients was 5.7±5.8 years (median 3.5 years; range: 1.5 months-32 years). Majority (61%) were males. Mean body weight was 15.4±11.6 kg (median 10.5 kg; range: 3-68 kg). Left ventricular end diastolic dimension (LVEDd) was 2.9±1.1 cm (range: 1.1-7.0 cm). Left ventricular ejection fraction (EF) was 62.3±7.5% (range: 35-80%). Sizes of main pulmonary artery (MPA), right pulmonary artery (RPA), and left pulmonary artery (LPA) were 10.4±3.8 mm (range: 0.75-25 mm), 8.5±3.3 mm (range: 1.25-20 mm), and 7.5±2.8 mm (range: 0.75-20 mm); respectively. Oxygen saturation (SpO2) on admission was 68.6±13.7% (range: 34-98%). Nineteen patients (5.8%) underwent BCPS as second stage palliation. Of those, 6 patients (1.8%) had undergone previous modified Blalock-Taussig shunt (m-BTS) operation, 5 patients (1.5%) had undergone modified central shunt (CS) operation, and 8 patients (2.4%) had undergone pulmonary artery banding (PAB). Of the 8 patients who had undergone PAB, one had undergone PAB plus atrial septostomy 4 months prior to BCPS. Of the 11 patients who had undergone m-BTS/CS, the shunt was patent in 8 patients. Most common primary lesion requiring BCPS was double outlet right ventricle (DORV) (30%), followed by tricuspid atresia (TA) (23%), transposition of great arteries (TGA) (14%), complex single ventricle (13%), tetralogy of Fallot (TOF) (7.3%), atrioventricular septal defect (AVSD) (5.5%). Eight patients had ventricular septal defect (VSD), 6 patients had pulmonary atresia (PA), 3 patients had Ebstein’s anomaly, 2 patients had hypoplastic tricuspid valve, and 1 patient had truncus arteriosus. Twenty-two different kinds of additional diagnoses, either alone or in combination, were present in 85 patients (26.1%) (Table 1).

Intraoperative and postoperative characteristics are summarized in Table 2. Majority of the patients (88%, n=286) underwent unilateral BCPS. Ninety-five percent (n=311) of the patients underwent the procedure by using cardiopulmonary bypass (CPB). In those patients who underwent BCPS with off pump technique, a temporary shunt was established between superior vena cava (SVC) and right atrium (RA) to decompress SVC during clamping. Mean CPB time was 80±47 min (range: 21-301 min). Six patients (2%) required institution of second pump run; with one of those requiring third time pump run. Aortic cross clamp (AoX) was applied whenever any intracardiac procedure had to be combined with the BCPS. Thirty-one percent (n=100) of the patients required the use of AoX. Mean AoX time was 30±23 min (range: 1-126 min). Two patients required second time AoX intraoperatively. Thirty-nine percent (n=128) of the patients required some kind of concomitant procedures along with BCPS. Twenty-three different concomitant procedures, either alone or in combination, were carried out in 127 patients. IAS excision (12%) was the most common concomitant procedure, followed by PA banding (11%), PDA ligation (10%), AVV repair (5%), MV repair (3%), and m-BTS/CS takedown (2%). Of the nine patients who underwent MV repair, annuloplasty ring was used in two patients (32 year old male, and 11 year old girl). Other concomitant procedures are presented in Table 2.

Figure 1: Chronology of landmark number of cases over years.
Table 1: Baseline Characteristics

| Variables                     | Overall patients (n=326) |
|-------------------------------|--------------------------|
| **Age, years; (mean±SD)**     | 5.7±5.8                  |
| **Sex, male; n (%)**          | 199 (61.0)               |
| **Weight (N=317), kg; (mean±SD)** | 15.4±11.6               |
| **LVDd (N=139), cm; (mean±SD)** | 2.9±1.1                 |
| **EF (N=131), (%); (mean±SD)** | 62.3±7.5                |
| **MPA size (N=99), mm; (mean±SD)** | 10.4±3.8                |
| **MPA z score (N=99); (mean±SD)** | -2.17±2.47              |
| **RPA size (N=114), mm; (mean±SD)** | 8.5±3.3                 |
| **RPA z score (N=114); (mean±SD)** | -0.44±2.02              |
| **LPA size (N=102), mm; (mean±SD)** | 7.5±2.8                 |
| **LPA z score (N=102); (mean±SD)** | -0.34±1.86              |
| **SpO2 on admission (N=89), %; (mean±SD)** | 68.6±13.7               |
| **Previous m-BTS/ CS/PAB; n (%)** | 19 (5.8)                |
| **Diagnosis/primary lesion, n (%)** |                              |
| DORV*                         | 99 (30.3)                |
| Tricuspid atresia             | 76 (23.3)                |
| TGA**                         | 45 (13.8)                |
| Complex single ventricle***   | 44 (13.5)                |
| TOF****                       | 24 (7.3)                 |
| AVSD*****                     | 18 (5.5)                 |
| VSD******                     | 8 (2.4)                  |
| PA                            | 6 (1.8)                  |
| Ebstein’s anomaly             | 3 (0.9)                  |
| Hypoplastic TV/TV anomaly     | 2 (0.6)                  |
| Truncus arteriosus            | 1 (0.3)                  |
| **Additional diagnosis**      |                          |
| Dextrocardia, n (%)           | 28 (8.6)                 |
| Situs inversus, n (%)         | 4 (1.2)                  |
| Ventricular inversion, n (%)  | 2 (0.6)                  |
| Situs ambiguous, n (%)        | 1 (0.3)                  |
| Mesocardia, n (%)             | 1 (0.3)                  |
| Right isomerism, n (%)        | 1 (0.3)                  |
| PDA dependency, n (%)         | 12 (3.6)                 |
| MR, n (%)                     | 9 (2.8)                  |
| RV dysfunction, n (%)         | 8 (2.4)                  |

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Azygos continuation of IVC, n (%) 5 (1.5)
Infective endocarditis, n (%) 3 (0.9)
PDA stenting, n (%) 2 (0.6)
Down’s syndrome, n (%) 2 (0.6)
LPA ostial stenosis 2 (0.6)
RPA ostial stenosis, n (%) 1 (0.3)
Post atrial septostomy, n (%) 1 (0.3)
TR, n (%) 1 (0.3)
AS, n (%) 1 (0.3)
AR, n (%) 1 (0.3)
ARCAPA, n (%) 1 (0.3)
CHB, n (%) 1 (0.3)
CRF, n (%) 1 (0.3)

Footnotes: * DORV PS 39, DORV AVSD II, DORV PA I, DORV TGA 40, DORV TGA AVSD 5, and DORV TGA P 3. ** TGA 7, TGA ASD PS 1, TGA PA 3, TGA PS 4, TGA VSD MA 1, TGA VSD PA 7, TGA AVSD PS 20, TGA AVSD 2. *** includes single ventricle with TAPVC. **** TOF 17, TOF PA 7. ***** AVSD 8, AVSD TGA 7, AVSD TGA TAPVC 1, AVSD TOF 1, AVSD TS 1. ****** VSD PS 5, VSD PA 4, VSD PAB 1.

N=number of data available for that particular variable. LVDd=left ventricular end diastolic dimension, EF=ejection fraction, MPA=main pulmonary artery, RPA=right pulmonary artery, LPA=left pulmonary artery, SpO2=oxygen saturation, m-BTS=modified Blalock-Taussig shunt, CS=modified central shunt, PAB=pulmonary artery banding, DORV=double outlet right ventricle, TGA=transposition of great arteries, TOF=tetralogy of Fallot, AVSD=atrioventricular septal defect, VSD=ventricular septal defect, PS=pulmonary stenosis, PA=pulmonary atresia, TV=tricuspid valve, ASD=atrial septal defect, MA=mitral atresia, TAPVC=total anomalous pulmonary venous connection, TS=tricuspid stenosis, PDA=pulmonary stenosis, RV=right ventricle, MR=mitral regurgitation, IVC=infant inferior vena cava, TR=tricuspid regurgitation, AS=aortic stenosis, AR=aortic regurgitation, ARCAPA=anomalous origin of right coronary artery from pulmonary artery, CHB=complete heart block, CRF=chronic renal failure, SD=standard deviation.

PA pressure was not routinely measured after the completion of anastomosis. In those cases, where it was measured (n=33), it was 18.6±5.1 mmHg (range: 10-30 mmHg). Central venous pressure (CVP) upon arrival to ICU was 19.2±4.6 mmHg (range: 6-30 mmHg). Duration of mechanical ventilation was 12.6±30.0 hours (median 6 hours; range: 40 min-309 hours). Duration of ICU stay was 3.9±4.8 days (range: 0-66 days). All patients who had ICU stay of zero days had actually mortality on the same postoperative day. Reexploration for bleeding or BCPS takedown was required in 11 patients (3.4%), of those 3 required BCPS takedown. Of the three cases requiring BCPS takedown, one was taken down in the same OT setting itself, one was taken down on the same night (postoperative day; POD 0), and the remaining one was taken down on POD 9. Extracorporeal
Variables Overall patients (n=326)

Operative procedure
- Unilateral; n (%) 286 (87.7)
- Bilateral; n (%) 40 (12.2)

Use of CPB
- Yes; n (%) 311 (95.4)
- No; n (%) 15 (4.6)

CPB time, min; (mean±SD) 80±47

Need for second pump run, yes; n (%) 6 (1.8)

Use of aortic cross clamp, yes; n (%) 100 (30.6)

AoX time, min; (mean±SD) 30±23

Need for second AoX, yes; n (%) 2 (0.6)

Concomitant procedures
- IAS excision; n (%) 40 (12.3)
- PAB; n (%) 35 (10.7)
- PDA ligation; n (%) 34 (10.4)
- AVV repair; n (%) 18 (5.5)
- MV repair; n (%) 9 (2.8)
- m-BTS/CS takedown; n (%) 8 (2.4)
- MPA closure/division; n (%) 4 (1.2)
- PA plasty/augmentation; n (%) 4 (1.2)
- TV repair; n (%) 4 (1.2)
- VSD enlargement; n (%) 3 (0.9)
- AV repair; n (%) 2 (0.6)
- LPA augmentation; n (%) 2 (0.6)
- TV closure; n (%) 2 (0.6)
- PAB tightening; n (%) 2 (0.6)
- ASD closure; n (%) 1 (0.3)
- Biopsy of vascular hemangioma of MV; n (%) 1 (0.3)
- Unifocalization of PA; n (%) 1 (0.3)
- RPA augmentation; n (%) 1 (0.3)
- PA separation; n (%) 1 (0.3)
- SVC-RPA junction augmentation with pericardium; n (%) 1 (0.3)
- Intrapulmonary PDA stent resection; n (%) 1 (0.3)
- Translocation of RCA; n (%) 1 (0.3)
- Epicardial pacemaker implantation 1 (0.3)

Mean PAP after anastomosis (N=33), mmHg; (mean±SD) 18.6±5.1

CVP upon ICU arrival (N=95), mmHg; (mean±SD) 19.2±4.6

Duration of mechanical ventilation (N=235), hours; (mean±SD) 12.6±3.0

ICU stay (N=294), days; (mean±SD) 3.9±4.8

Hospital stay (N=126), days; (mean±SD) 7.8±5.9

Reexploration, yes; n (%) 11 (3.4)

BCPS takedown, yes; n (%) 3 (0.9)

Use of ECMO, yes; n (%) 2 (0.6)

Reintubation, yes; n (%) 8 (2.5)

Inotropes (N=227)
- Single inotrope; n (%) 62 (27.3)
- Two inotropes; n (%) 72 (31.7)
- Three inotropes; n (%) 71 (31.3)
- Four or more inotropes; n (%) 22 (9.7)

Mortality; yes; n (%) 49 (15)

ICU readmission, yes; n (%) 8 (2.5)

Chylothorax, yes; n (%) 7 (2.1)

Phrenic nerve palsy; n (%) 2 (0.6)

SpO2 on discharge (N= 83), %; (mean±SD) 79.2±9.5

Change in SpO2 % at the time of discharge from baseline value (ΔSpO2) (N=70), %; (mean±SD) 10.7±13.3

Footnotes: N=number of data available for that particular variable, CPB=cardiopulmonary bypass, AoX=aortic cross clamp, IAS=interatrial septum, PAB=PA banding, PDA=patent ductus arteriosus, AVV=atrioventricular valve, MV=mitral valve, m-BTS=modified Blalock-Taussig shunt, CS=modified central shunt, MPA=main pulmonary artery, PA=pulmonary artery, TV=tricuspid valve, VSD=ventricular septal defect, AV=aortic valve, LPA=left pulmonary artery, ASD=atrial septal defect, PAP=pulmonary artery pressure, CVP=central venous pressure, ICU=intensive care unit, BCPS=bidirectional cavopulmonary shunt, ECMO=extracorporeal membrane oxygenation, ΔSpO2=change in oxygen saturation, SD=standard deviation.

Membrane oxygenation (ECMO) had to be instituted in two patients. Eight patients (2.5%) required ICU readmission. Reintubation was required in 8 patients (2.5%). Postoperative chylothorax was noted in 7 patients (2.1%) of which 4 patients required thoracic duct mass ligation, 2 patients required chemical pleurodesis and octreotide, and remaining one patient responded with octreotide alone. Phrenic nerve palsy was observed in 2 patients (0.6%), of which it was bilateral.
in one patient. One of those two patients required diaphragmatic plication.

In hospital mortality was 15% (n=49). Mortality was due to low cardiac output syndrome (LCOS) in 22 patients (44.9%), intractable VT/VF in 2 patients (4.1%), multiple organ dysfunction syndrome (MODS) in 2 patients (4.1%), sepsis in 1 patient (2%), and hypoventilation due to blockage of tracheostomy tube in 1 patient (2%). In 21 patients (42.8%), the cause of the death was not available. Of those who died, more than half (n=26) died within 2nd postoperative day (POD 2) (Figure 2). One of those deaths occurred in the OT itself due to failure to wean from bypass. Patients who died had low body weight (11.8±10.7 kg vs 16.0±11.6 kg; p=0.019), longer CPB time (101±64 min vs 76±42 min; p=0.001), and these patients tended to be younger although statistically not significant (4.6±7.0 years vs 5.9±5.5 years; p=0.108). Duration of postoperative hospital stay for patients who were discharged home was 7.8±5.9 days (range: 3-41 days).

Mortality was 15% (n=49). Most of the mortality has been reported on postoperative day 1 (POD 1).12,13 First clinical application of cavopulmonary shunt was described by Shumacker HB in 1955.12 Dr William WL Glenn from Yale University further popularized this procedure with his experimental work in dogs and subsequent translation into clinical practice.13,14 and today this procedure bears his name on it as Glenn procedure (classic Glenn/ bidirectional Glenn).

First case described by Glenn was performed via right thoracotomy approach in Feb 25, 1958 in a 7-year-old boy with single ventricle, transposition of great arteries, and pulmonary stenosis.14 Immediately after the operation, cyanosis almost disappeared. Two months after surgery, SpO2 at rest increased to 86.9% from 70.7% prior to surgery. Also, hematocrit decreased from 70% to 47%. The most striking change after operation was the increase in exercise tolerance.14 Glenn and his team described its clinical application to 38 cases after 6 year of first successful SVC to PA anastomosis;12 where the authors argued that the anastomosis shows growth potential as the adjacent vessels grow; and superior caval pressure remains moderately elevated for at least several years after the procedure, as shown by the brachial venous pressure measurements. Half a century experience after classic Glenn has recently been described by a group from Yale.16

In their analysis, a patient who had undergone classic Glenn for tricuspid atresia at the age of 6 months was still alive at the age of 54 years. Over time, the classic Glenn shunt, where superior caval flow is directed only to the right lung, has evolved to a unidirectional to a bidirectional cavopulmonary shunt (BCPS) which provides less distortion to the right pulmonary artery and allows for blood flow to both the lungs. Since early 1980s, bidirectional anastomosis, where transected end of SVC is anastomosed to right pulmonary artery in an end-to-side fashion without transecting the proximal right PA, was popularized.17 Unlike in classic Glenn shunt, the continuity of right and left pulmonary arteries is maintained in BCPS, thereby allowing further completion of Fontan procedure at a later date.

In our center, we have never performed classic Glenn anastomosis. We performed our first case of BCPS on a 7-year-old boy with tricuspid atresia on February 7, 2002 (2058/10/25 BS). It is of a nice coincidence that the first clinical case of cavopulmonary anastomosis done by Dr Glenn was also performed on the month of February and on a 7-year-old boy.14 We performed our 50th case six-and-half years later on July 29, 2008. Landmark for 100th case was reached on Dec 10, 2010. Our landmark dates have been presented in Figure 1.

Young age, low body weight has been described as predictors of in hospital mortality previously.2,4-9 Although the patients who died in our series tended to be younger, no statistically significant difference was noted for age as opposed to body weight, where the patients who died had significantly low body weight (p=0.019). Most common primary lesion requiring BCPS was DORV, followed by tricuspid atresia, TGA and others; which was quite similar to the findings of other authors.15,16,17 Although we had many patients who had undergone BCPS as a part of one and half ventricul repair for Ebstein’s anomaly, we excluded those during analysis because we believe this group of population deserves a separate analysis. In this series, we had only 3 patients who had undergone isolated BCPS for Ebstein’s anomaly without tricuspid repair.

One of the major advantages of BCPS is to decrease the volume overload of systemic ventricle/ right ventricle thereby preventing right ventricular failure. In our series, eight patients already had

Discussion

Anastomosis between superior vena cava and right pulmonary artery to palliate certain congenital heart diseases with decreased pulmonary blood flow as a way of bypassing the right ventricle has been described as early as 1950s.11 This procedure was developed by several surgical groups working independently and probably unaware of each other ranging from Italy to Soviet Union to the United States.11

SpO2 in room air at the time of discharge was 79.2±9.5%; which was significantly higher than that of SpO2 in room air at the time of admission (68.6±13.7%) (p=0.000). Mean SpO2% change at the time of admission (ΔSpO2) was significantly higher than that of SpO2 in room air at the time of admission (68.6±13.7%) (p=0.000). Mean SpO2% change after operation was the increase in exercise tolerance.14 Glenn and his team described its clinical application to 38 cases after 6 year of first successful SVC to PA anastomosis;12 where the authors argued that the anastomosis shows growth potential as the adjacent vessels grow; and superior caval pressure remains moderately elevated for at least several years after the procedure, as shown by the brachial venous pressure measurements. Half a century experience after classic Glenn has recently been described by a group from Yale.16

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Figure 2: Bar diagram showing mortality and postoperative day.

Most of the mortality has been reported on postoperative day 1 (POD 1).

Figure 3: Comparison between preoperative and postoperative

SpO2 in room air for individual patients. Most of the patients had their SpO2 levels increased at the time of discharge except for few who had lower SpO2 levels at discharge time than that of admission time.

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Because presence of residual A VVR leads to poor outcomes, we always take down the previously created m-BT or central shunts. There have been reports of veno-venous collaterals between SVC into lungs and subsequent desaturation. Even after ligation of azygos veins, there is presence of bilateral SVC, we always perform bilateral BCPS.

In our series, 12% of the patients underwent bilateral BCPS. We do not routinely perform catheterization studies to find out the physiological status of pulmonary vasculature. Pulse oximetry data both at the time of admission and discharge was available only in 70 patients. Of those 70 patients, majority had their SpO2 levels increased at the time of discharge as compared with the time of admission; except 12 patients (17.1%) who had lower SpO2 levels at the time of discharge as compared to the time of admission. This might probably be attributable to the poor chest physiotherapy and lung atelectasis/collapse. We do not have precise numerical data of how all these patients did in follow up; however, most of the kids in this category showed marked improvement of symptoms even with low saturations at the time of discharge and on OPD follow ups.

**Limitations**

Because of the retrospective design of the study, it carries inherent flaws related to retrospective nature. Because we could not obtain all variables dated back to 18 years prior, not every variable has the n of 326. We have mentioned only the available numbers as N after each variable in the two tables presented. Because this study focuses only on in-hospital outcomes, follow up data is not available.

**Conclusion**

This is the first report of its kind to analyze surgical outcomes of BCPS in our center. We have discussed the history and evolution of BCPS surgery in our center. We have performed this surgery for a variety of congenital heart lesions and have shown the outcomes in terms of morbidity and mortality. Our in hospital mortality remains still high; and we believe proper patient selection prior to the decision of BCPS surgery is utmost important and should be the way forward. We believe we will have to make a protocol of doing BCPS in our center for patient selection which includes preoperative cardiac catheterization mandatory for every case.

**Conflicts of interest / disclosures:** None

**Acknowledgement:** None

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