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Research Article

Palliative Shunt Versus Ductal Stenting in Duct -Dependent Pulmonary Circulation

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Abstract

Background: Neonatal surgical palliative systemic to pulmonary artery shunt is still a major cause of mortality and morbidity. Recently, ductal stenting (DS) is an effective alternative modality to provide adequate blood flow in neonatal congenital heart disease with duct-dependent pulmonary circulation. The aim of this study was assess feasibility and outcomes of DS in comparison to a conventional modified Blalock-Taussig shunt (MBTS) with duct-dependent pulmonary circulation.

Methods: A single-institution retrospective cohort study was conducted in neonates who underwent MBTS and DS. Analysis of survival, procedural complications, rate of re-stenosis and re-intervention were done. Log rank (Mantel-cox) was done to compare the means and median re-stenosis survival time.

Results: A total of 67 subjects ≤ 12-month-old were identified. Eighteen subjects underwent DS and 49 subjects underwent MBTS. The length of stay and overall hospital mortality were similar. Pneumonia intra-hospital and sepsis after procedure were higher in MBTS group. The mean and median survival time for re-stenosis of DS and MTBS was 25.32 and 77.82 months (p = 0.001) respectively, however the rate of re-intervention was not different.

Conclusion: DS and MBTS in aged < 6 months are feasible and may provide an effective systemic to pulmonary artery shunt in duct-dependent pulmonary circulation in our institution, however DS efficacy gradually reduced.

Keywords: *Palliative Shunt; Ductal Stenting; Duct-Dependent Pulmonary Circulation*

Background

Systemic-to-pulmonary artery (S to PA) surgical shunting is commonly used as a bridge to the next palliative stage or definitive surgical repair in patients with duct-dependent pulmonary circulation during early infancy period including the patient with single ventricle or two ventricles who have severe pulmonary stenosis or atresia [1].

However, although several modifications of surgical technique and post-operative managements have been made, the mortality and morbidity of surgical shunt procedure in early stage of life remains notably high [2,3].

Ductal stenting (DS) is recently reported as an effective alternative modality to establish adequate pulmonary blood flow in patients with duct-dependent pulmonary circulation [4,5]. The results of DS varies among institutions [6,7]. Some difficulties or limitations of DS procedures are met in neonates related to small patient size and duct morphology. Procedural failure appeared in approximately 10% of

cases and usually occurred in the tortuous ductus arteriosus whilst the morbidity and mortality was 8 - 11% and less than 1%, respectively [8].

However, there is limited data about whether percutaneous ductal stenting approach could be the alternative to conventional surgical treatment. This study was to compare the results of surgical conventional systemic to pulmonary artery shunt and percutaneous ductal stenting in cyanotic congenital heart disease (CCHD) newborns or infants with ductal-dependent pulmonary circulation.

Methods

We performed a retrospective study of the patients who underwent SP shunt (modified Blalog Tausig shunt/MTBS) and DS at Queen Sirikit National Institute of Child Health, Bangkok, Thailand from January 2010 to December 2014. Local ethics committee has approved this study. The inclusion criteria were patients aged less than 6 months old, pulmonary duct dependent. Patients underwent corrective surgery were excluded from the study. Medical records were reviewed for the patient's characteristics, pre-procedural status, details of the procedure, post-operative course, morbidity, and mortality.

Patient's characteristics included gender, birth weight, presence of significant non-cardiac congenital abnormalities, and the diagnosis of CCHD. Prematurity was defined as a gestational age at birth of less than 37 completed weeks of gestation. Low birth weight was defined categorically as less than 2.5 kg. The diagnosis of CHD was defined by reviewing of the information from echocardiogram, cardiac catheterization, and operation. In case of disagreement of the diagnosis, listed in the operative report was used. Selection of patients for MTBS and DS were chosen from a cardiac discussion.

Cardiac lesions were categorized according to shared anatomic and physiologic characteristics: (1) tricuspid atresia and variants, (2) pulmonary atresia with intact ventricular septum (PA-IVS) and variants (3) tetralogy of Fallot (ToF) or pulmonary atresia with ventricular septal defect (PA-VSD) and variants, (4) other functional single ventricle lesions with obstruction to systemic or pulmonary blood flow.

Intra- and post-operative variables related to MTBS or DS including type and diameter of the implanted shunt or stent usage of cardiopulmonary bypass, extracorporeal membrane oxygenation (ECMO) use, laboratory parameters, usage of heparin infusion in the post-operative period, number of bloodstream or surgical wound infections, duration of hospital stay, and mortality. Mortality was classified as early (occurred within 30 days after procedure) or late (more than 30 days after procedure). Procedural related infection was defined as distinct clinical episodes requiring intravenous antibiotics therapy along with positive blood or surgical wound cultures during hospital stay.

The mechanism of shunt or stent dysfunction leading to re-intervention was ascertained by echocardiogram, angiogram, or operative report. It was classified as secondary to thrombosis, mechanical distortion of the shunt or pulmonary artery, combination of thrombosis and distortion, or indeterminate.

At follow-up, Changes of the indexed sum of the cross sectional pulmonary artery areas (Nakata index) and the ratio of the sum of the diameters of both pulmonary arteries to the diameter of the descending aorta (McGoon ratio) as well as the right and left pulmonary artery (LPA) Z-scores will be evaluated.

Statistical analysis was performed with SPSS. Data are reported as mean (standard deviation) for normally distributed data or median (range) otherwise. Categorical data are presented as n (%). Analysis of survival, procedural complications, rate of re-stenosis and re-intervention were done. Log rank (Mantel-cox) was done to compare the means and median re-stenosis survival time. Significance was defined as $p < 0.05$.

Results

During the study period, 95 patients underwent systemic to pulmonary shunt. Twenty eight patients were excluded which included 7 cases of cyanotic CHD with duct dependent systemic circulation, 21 cases who had age more than 1 year at the procedure. As a result, 49 systemic to pulmonary shunt (MBTS) patients and 18 patients who underwent DS were enrolled into this study during the time period. Pre-procedural patient's characteristics were demonstrated in table 1.

Patients	SP shunt (n = 49)	DS (n = 18)
Preterm, n (%)	1 (2)	3 (16.7)
Age at intervention, median (range)	5 days (3 days to 2.5 months)	15 days (3 days to 5.22 months)
Weight at intervention (kg), median (range)	3.2 (2.3 to 7.2)	3 (2.25 to 10.8)
Sex, n (%)		
Male	28 (57.1)	10 (55.6)
Saturation (%), mean, (SD)	73.8 (8.5)	71.3 (11.4)
Diagnosis		
TA and variants	2 (4.1)	0
PA, VSD	13 (26.5)	10 (55.6)
PA, IVS	6 (12.2)	1 (5.6)
Tetralogy of Fallot	8 (16.3)	0
Single ventricle	2 (4.1)	0
Isomerism	5 (10.2)	4 (22.2)
PVS	0	3 (16.7)
Complex CHD	13 (26.5)	0
PGE1 Infusion		
Yes, n (%)	33 (67.3)	14 (77.8)
Duration (days), mean (SD)	7.5 (10.22)	8.36 (8.4)
RPA (mm), median (range)	3.8; 2-5	3.9; 3.1-9.4
RPA z-score, median (range)	-0.17, (-0.84; 1.03)	0.04, (-0.78; 2.70)
LPA (mm), mean (SD)	3.72 (1.07)	4.57 (2.36)
LPA z-score, mean (SD)	-0.11 (0.67)	0.16 (1.12)
Nakata Index (mm^2/m^2), mean (SD)	128.5 (40.92)	139 (55.64)
McGoon Ratio, mean (SD)	1.28 (0.27)	1.33 (0.27)

Table 1: *Pre-procedure patient 's characteristics.*

Abbreviations: kg: kilogram; PGE1: Prostaglandin-E1; TA: Tricuspid Atresia; PA, VSD: Pulmonary Atresia, Ventricular Septal Defect; PA, IVS: Pulmonary Atresia, Intact Ventricular Septum; PVS: Pulmonary Valvular Stenosis; RPA: Right Pulmonary Artery; LPA: Left Pulmonary Artery; SD: Standard Deviation.

In systemic to pulmonary shunt group, MBTS was performed in all the patients. The MBTS was mostly operated with a standard postero-lateral thoracotomy from the fourth intercostal space incision, only one patient with ToF, right aortic arch and left pulmonary artery stenosis was performed from median sternotomy. The MBTS was undertaken from the right side in 83.7% and 14.3% from the left side. Polytetrafluoroethylene (PTFE) graft was used in all MBT shunts with the mean diameter was 4.0 (SD 0.36) millimeters.

In DS group, cardiac catheterization was performed under general anesthesia in all the patients with the mean procedural time was 100 (SD 43.9) minutes and the mean fluoroscopy time was 35.14 (SD 2.68) minutes. Stent implantation was undergone from various approaches depending upon the origin and morphology of the arterial duct in relations to the underlying cardiac defects. Stenting of the duct were performed from the femoral vein across the ventricular outflow tract and femoral artery. Ductus arteriosus types were elongated arterial duct originating from descending aorta in 9/18 patients, slightly curved arterial duct originating from the distal aortic arch (U type) in 7/18 patients, and tortuous arterial duct originating from the aortic arch in 2/18 patients. Balloon expandable coronary stents were used such as Prokinetic (Biotronika AG, Bulach, Switzerland) in 72.2% patients and Multilink Vision (Abbot vascular, Illinois, USA) in 27.8% patients. The mean diameter of the stent was 4.36 (SD 0.37) millimeters and the length was 14.39 (SD 1.6) millimeters.

Pulmonary valvuloplasty and balloon atrial septostomy were additionally performed in 4 patients. Procedure-related complications occurred in 4/49 of SP shunt and 7/18 in DS group (Table 2). No procedure-related mortality was found in both groups.

	SP shunt (n = 49)	DS (n = 18)
Additional procedures, n (%)		
PBV	1 (2)	3 (16.7)
BAS	0	1 (5.6)
Procedure-related complications, n (%)		
Thrombosis	3 (6.1)	0
Migration stent	0	3 (16.7)
Pulmonary hemorrhage	0	1 (5.6)
Uncovered	0	3 (16.7)
Seroma	1 (2)	0
Success rate (%)	91.8	83.3

Table 2: *Additional procedures and immediate complications*
PBV: Percutaneous Balloon Valvuloplasty; BAS: Balloon Atrial Septostomy.

During hospitalization, there was no statistical difference of the presence of infections, hospital length of stay, in-hospital complications and mortality between the two groups. However, the number of patients with pneumonia was higher in MTBS group (Table 3).

	SP shunt (n = 49)	DS (n = 18)	P value
Presence of infection, n (%)	30 (61.2)	12 (66.7)	0.289
Hospital length of stay (days), mean (SD)	22.48 (14.98)	21.44 (14.18)	0.801
In-hospital complications, n (%)			
Sepsis	16 (32.6)	7 (38.9)	0.002
Pneumonia	13 (26.5)	1 (5.6)	
Heart failure	0 (0)	2 (11.1)	
Others*	4 (8.2)	6 (33.3)	
No	20 (40.8)	2 (11.1)	
In-hospital survival, n(%)			
Survived	43 (87.7)	15 (83.3)	0.753
Early mortality	6 (12.2)	3 (16.7)	

Table 3: *In-hospital conditions after procedure.*
Others: meningitis, atelectasis (in SP shunt), coagulopathy, stroke, brain edema (in DS).

Follow-up data was achieved in 55 patients with the mean duration of follow up was 18.03 (11.04) months in MTBS group and 13.75 (7.04) months in DS group (Table 4). Re-intervention was not different between 2 groups. In one subject, re-intervention at group II required SP shunt urgent due to recurrent cyanosis. Late re-intervention or second shunt was created in SP shunt group such as L-BT shunt in 3 patients, Central Shunt in 1 patient and MAPCA coil in 1 patient and in DS group was BPV with BAS in 1 patient. PA branches stenosis rate were higher in SP group such as RPA stenosis, LPA stenosis, PA bifurcation stenosis, and combination of RPA and LPA stenosis.

	SP shunt (n = 40)	DS (n = 15)	P value
Duration of follow up (months), mean (SD)	18.03 (11.40)	13.75 (7.04)	0.564
Oxygen saturation (%), mean (SD)	79.88 (6.9)	71.50 (8.18)	0.791
RPA (mm), mean (SD)	8.84 (3.06)	11.47 (4.18)	0.247
RPA z-score, mean (SD)	-0.006 (1.05)	0.89 (1.44)	0.246
RPA before –RPA after, mean (SD)	4.60 (2.3)	5.77 (4.35)	0.484
LPA (mm), mean (SD)	9.73 (2.76)	7.5 (0.53)	0.136
LPA z-score, mean (SD)	0.43 (1.11)	-0.46 (0.21)	0.123
LPA before –LPA after, mean (SD)	5.99 (2.78)	2.3 (1.48)	0.078
Nakata Index (mm^2/m^2), mean (SD)	314 (109)	381 (204.70)	0.513
% Increase**			
McGoon Ratio, mean (SD)	2.09 (0.43)	2.35 (0.64)	0.756
% Increase**			
Re-stenosis, n (%)	2 (4.1)	7 (38.9)	< 0.001
Re-intervention, n (%)	7 (14.3)	4 (22.2)	0.383

Table 4: *Follow-up data.*

The incidence of shunt/stent re-stenosis at follow up was significantly higher and earlier in patients after DS procedure ($p < 0.001$). Moreover, once re-stenosis occurred, the survival rate was significantly decreased in DS group. Rate of re-stenosis in DS group was higher than SP group (Table 4) and from survival chart shown re-stenosis was earlier in DS group (Figure 1).

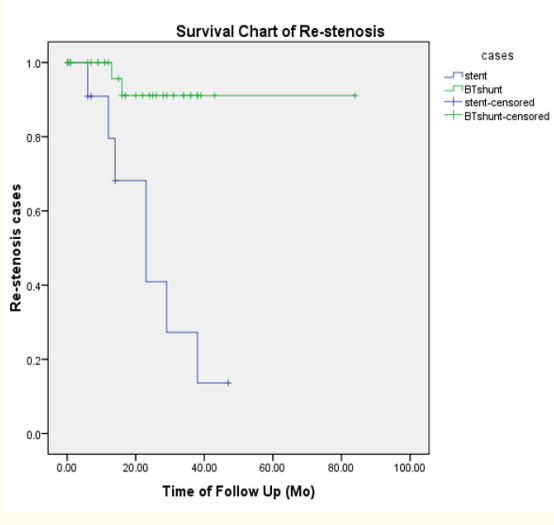

Figure 1: *Survival rate of the patients with re-stenosis of SP shunt and DS ($p < 0.0001$).*

Discussion

Historically, BT shunt was considered the primary management option for ToF. In the current era, thus SP shunts are more commonly indicated and as the first line palliative treatment for infants with complex single-ventricle physiology [2,3]. The various diagnosis in this

study for which group 1 or 2 was performed, such as with single ventricle or complex congenital heart disease in group BT shunt, however the most frequent diagnosis in both groups were PA, VSD.

The majority of patients underwent DS was younger than MTBS in this study, which is at an early age ranging between 3 days to 5 months. Aged at intervention influences the outcomes. In newborns with pulmonary atresia, ductal closure during MBTS procedure is associated with increased incidence of resuscitation events, need for re-intervention, and increased mortality during the early postoperative period [9].

Successful rate of DS procedure is varies in many institutions. A multicenter experience over 18 years reported DS procedure was successful in 80% neonates and infants and low incidence of freedom from re-intervention [6]. Procedural failure depends mainly on ductal tortuosity and ranges around 10% of cases. Morbidity and mortality are 8 - 11% and less than 1%, respectively [8]. Situation in our study showed mortality rate in DS group were higher, but the rate was similar with MTBS group. However, higher mortality rate than others may cause by intra-hospital complications such as high incidence of sepsis, pneumonia, or meningitis. Other institutions report such as Kuala Lumpur, they have been reported successful rate of DS which was 91.1%, no procedure-related mortality, and the freedom from re-intervention was 89% and 55% at 6 months and 1 year [7]. Otherwise, higher incidence of successful procedure reported in Istanbul, the stenting procedure was successfully completed in 93.3% [10].

Comparing outcomes of SP and DS remain varies in some institutions. The sizes of RPA and LPA or Z score of both PA branches were similar after procedure. Mc.Goon and Nakata index were increase in both groups with similar result. Some studies revealed after 10 (SD 5) months, both options had promoted a significant increase of the Nakata index and McGoon ratio [11,12]. However, there were a greater percentage of shunted patients experienced procedure-related complications and distal branch pulmonary artery stenosis, and this result similar with other study [13].

There was higher procedure-related complications in DS group such as migration/protrusion of the stent, uncovered stent, and pulmonary hemorrhage. Protruding stents do not cause an increased risk of thrombosis in patients on aspirin. There is no statistical evidence that protrusion can cause lung perfusion defects [14]. Furthermore, migration of the stent is not a life-threatening complication, but nonetheless serious as the patient needs to go to surgery semi-electively for stent removal and construction of a MTBS. With proper case selection, appropriate technique, and the right hardware ductal stenting provides reasonable short-medium term palliation in duct-dependent cyanotic heart disease [15].

The most feared complication is fatal shunt thrombosis. At MTBS group there was thrombosis as complications but none in DS group, while the early mortality noted was not differ. One might speculate that an insidious reduction in the shunt lumen size as a result of accumulation of platelet thrombi may produce a gradual decline in baseline oxygen saturation. Any stressful event, such respiratory or gastrointestinal disease, may precipitate an irreversible degree of hypoxemia and acidosis. Constant seeding of emboli into the systemic and pulmonary circuits is another possible mechanism of death [16]. Otherwise, the condition of the patient at the time of surgery and underlying cardiac pathology are influence the mortality [3].

Re-stenosis and re-intervention

Rate of re-stenosis was higher in DS group but longer in duration. Therefore, re-stenosis of DS could be more problematic and may be a disadvantage. Successful DS depends on the morphology of the duct as mentioned at AHA which is straight duct and who require reliable palliation for no more than 3 to 6 months are recommended for stenting [4,5]. One study in Egypt have been proven that efficacy of DS gradually lessens after 6 months due to intra-stent endothelial hyperplasia [17].

The incidence of surgical or catheter-based re-intervention to maintain adequate pulmonary blood flow was similar in both groups. This result was not different with other study [13], although DS also have several limitations, ductal stenting is preferable now in some centers with crowded surgical lists, indicated mainly in duct-dependent cyanotic lesions chiefly in the neonatal period, and sometimes the pulmonary arteries are too small for SP shunt [4,5].

Conclusion

DS and MBTS in aged < 6 months are feasible and may provide an effective systemic to pulmonary artery shunt in duct-dependent pulmonary circulation in our institution, however DS efficacy gradually reduced.

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