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Predicting Factors of Mortality in Patients after The Modified Blalock-Taussig Shunt Procedure in Developing Country : A Retrospective Study

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Abstract

Introduction: Blalock Taussig (BT) is a palliative procedure that preserves blood circulation to the lungs and alleviates cyanosis in patients with congenital heart diseases and reduced pulmonary blood flow. BT shunt remains a routinely performed procedure in developing countries before definitive surgery. However, evidence on predictors factors of mortality after this procedure is still scarce in Indonesia.
Aim: to evaluate the predictive factors of mortality after the BT shunt procedure.
Methods: This retrospective study evaluated the medical record data of all postoperative BT shunt patients at Dr. Cipto Mangunkusumo Hospital, Jakarta, Indonesia, from 2016 to 2020. We performed univariate and multivariate analyses to identify the predictors of in-hospital mortality.
Results: The total subjects in this study were 197 children, 107 (54.3%) boys and 90 (45.7%) girls. The median values for age and body weight at the time of surgery were 20 months (11 days – 32 years) and 7.9 (2.7 - 42) kg. The most prevalent diagnosis was the Tetralogy of Fallot, which was found in 80 (40.6%) patients. In-hospital postoperative mortality was 20.8% (41 patients). Based on multivariate analysis, predictors associated with mortality were weight <4.25 kg (OR 20.9; 95% CI 7.4-59.0; p <0.0001) and emergency procedures (OR 3.5; 95% CI 1,3-9.5; p =0.016).
Conclusion: The mortality rate after BT shunt at PJT Rumah Sakit Cipto Mangunkusumo was 20.8%. Based on multivariate analysis, weight <4.25 kg and emergency procedures are two predictors of mortality in BT shunt.

Keywords: Blalock Taussig shunt, Congenital heart disease, Indonesia, Mortality

Introduction

Congenital heart disease (CHD) are the most common type of birth defects found in 8 per 1,000 live births.[1] In developing countries, most CHD are not detected until postnatal or later in life, during childhood or some until adulthood. About 25% of babies born with heart abnormality has critical CHD. These babies need surgery or any other procedure in the first year of life.[2]
Blalock Taussig (BT) shunt is one of the most common palliative procedures performed in CHD patients. BT shunt aims to improve blood flow to the pulmonary circulation, alleviate cyanosis, promote pulmonary artery growth, and preserve cardiac preload, afterload, and coronary arteries perfusion.[3] BT shunt is still commonly performed in developing countries before the definitive correction. But it has been rarely performed in developed countries as the development of surgical techniques, adequate technologies, the availability of experts have made it possible to perform the definitive surgery as early as possible.[4,5]
Mortality after BT shunt in Indonesia is still high, ranging from 12.5 to 14.6%.[6]Even though BT shunt has become an integral part of CHD treatment in Indonesia for years, research assessing its mortality is still scarce. Investigating BT shunt procedure outcomes and factors that affect them is crucial to measure and improve the quality of care in the future. Accordingly, this study aims to assess the predictive factors of mortality after the BT shunt procedure in Jakarta, Indonesia.

Methods

Study design and population

This retrospective cohort study was conducted at Dr. Cipto Mangunkusumo Hospital, Jakarta, Indonesia, from November 2020 to January 2021. During this period, all CHD pediatric

patients with a previous history of BT shunt procedure were included in the study. Data were primarily obtained from medical records from January 2016 to December 2020; thus, patients with incomplete records were excluded.

This study had been approved by the Ethics Committee of the Faculty of Medicine, University of Indonesia. The permission to assess the medical records was obtained from Dr. Cipto Mangunkusumo Hospital, Jakarta, Indonesia. Informed consent was obtained from the patients before participating in the study.

Data collection

We collected the baseline characteristics of patients' age, sex, weight, nutritional status, and diagnosis. Meanwhile, additional predictors of mortality after the BT shunt procedure assessed in this study were the type of surgery (urgency/emergency), surgical approaches (sternotomy or thoracotomy), and the need for packed red cell (PRC) transfusion after the procedure. We diagnosed malnutrition in children based on World Health Organization (WHO) growth chart 2006(Z score <-2 SD for children below 5 years old and Centers for Disease Control and Prevention (CDC) growth chart 2000 for children age 5-18 years old (weight/height <90%). These classification has been used by Indonesian Pediatric Association (IDAI).

Outcome measures

The primary outcome investigated was in-hospital mortality. In addition, we also found the predictor factors of mortality after the BT shunt procedure.

Data analysis

Data analysis was performed using IBM SPSS version 25.0. Categorical data were described with their absolute (n) and relative frequencies (%). For normally distributed quantitative data, we presented it with mean and standard deviation. On the other hand, we expressed them as median and range if the data was not normally distributed. The association between variables was assessed with bivariate analysis (chi-square or Fisher exact tests) initially. If the predictors had a p-value <0.25, they were deemed eligible for multivariate analysis, further determining the relationship between predictors and outcomes. The multivariate analysis results were presented as odds ratio (OR) and confidence interval (CI). P-value <0,05 or CI not including 1 was considered statistically significant.

Ethics approval and consent to participate

The Ethics Committee of the Faculty of Medicine, University of Indonesia – Cipto Mangunkusumo Hospital approved this study (KET-1301/UN2.F1/ETIK/PPM.00.02/2020).

Results

One hundred and ninety-seven patients met the inclusion and exclusion criteria of this study. The baseline characteristics are described in Table 1. Most of the subjects in this study have normal nutritional status. The most common diagnosis in this study was Tetralogy of Fallot (40,6%).

Table 1

Baseline Characteristics of The Study Subjects

Characteristics	n = 197
Sex, n (%)	
Men	107 (54.3)
Women	90 (45.7)
Weight, median (range) Kg	7.90 (2,7 - 42)
Age, median (range)	20 months (11 days – 32 years)
Nutritional status, n (%)	

Severe malnutrition	31 (15.7)
Mild-moderate malnutrition	48 (24.4)
Normal	93 (47.2)
Overweight/obese	25 (12.7)
Diagnosis, n (%)	
PA-IVS	18 (9.1)
PA-VSD	52 (26.44)
Tetralogy of Fallot	80 (40.6)
Ebstein Anomaly	1 (0.5)
CAVSD + PS	10 (5.1)
Tricuspid atresia	15 (7.6)
Mitral atresia	6 (3)
DOLV + PS	2 (1)
DORV + PS	7 (3.6)

Note. PA-IVS = pulmonary atresia with intact ventricular septum, PA-VSD = pulmonary atresia with ventricle septal defect, CAVSD = complete atrioventricular septal defect, PS = pulmonary stenosis, DOLV = double outlet left ventricle, DORV = double outlet right ventricle.

Table 2 shows the characteristics of the BT shunt procedure performed at our center. Most of the surgeries performed were elective (81.2%) and without cardiopulmonary bypass (86.3%). The most frequent shunt size was 4 mm, and the median diameter and weight ratio were 0.4 mm/Kg. The majority of subjects did not get blood transfusion after the procedure (58.4%).

Table 2.

Characteristics of Bt Shunt Procedures

Characteristics	n = 197
Type of surgery, n (%)	
Urgency/emergency	37 (18.8)
Elective	160 (81.2)
Surgical approach, n (%)	
Median sternotomy	90 (45.7)
Thoracotomy	107 (54.3)
CPB use, n (%)	
Yes	27 (13.7)
No	170 (86.3)
Graft size (mm), n (%)	
3	4 (2)
3.5	29 (14.7)
4	105 (53.3)
5	47 (23.9)
6	12 (6.1)
Ratio of Shunt diameter/weight, median (range) mm/Kg	0.4 (0.09 – 1.0)
Shunt position, n (%)	
Left Modified BT shunt	33 (16.8)
Right Modified BT shunt	119 (60.4)
Central shunt	45 (22.8)
Postoperative transfusion, n (%)	
Yes	82 (41.6)

No	115 (58.4)

*Note.*CPB = cardiopulmonary bypass

There were two numeric variables assessed separately from other categorical predictors, age and weight. We used the receiver operating characteristics curves to determine these variables' cut-off in predicting the mortality after the BT shunt procedure (Figure 1). From the curve, we found that the area under curve (AUC) value of weight as a mortality predictor was 81% (95% CI, 74.1 to 89.7%; p <0,0001). We determined the optimal weight's cut off from the line curve to predict the mortality after BT shunt, which was 4.25 Kg (sensitivity: 51.2%; specificity: 94.2%) (Figure 2). Meanwhile, the AUC value for age was 75% (95% CI, 66.9 to 84.9%; p <0,0001) (Figure 3). The age's cut-off for predicting mortality was 270 days (sensitivity: 61%; specificity: 78.2%) (Figure 4)

Figure 1.

ROC Curve of Weight as A Predictor for Mortality After BT Shunt.

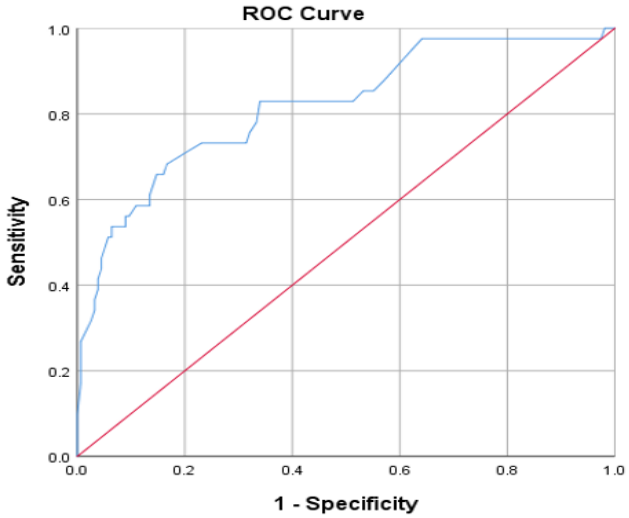


Figure 2

Weight Cut-off in Predicting Mortality

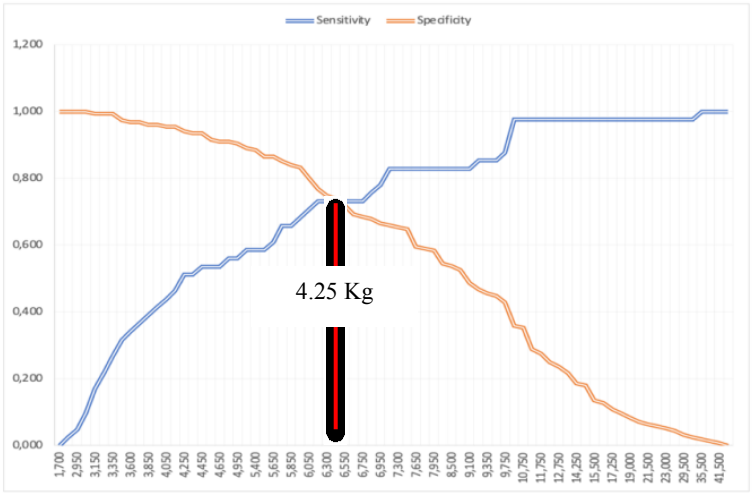


Figure 3

Roc Curve of Age As A Predictor for Mortality after BT Shunt

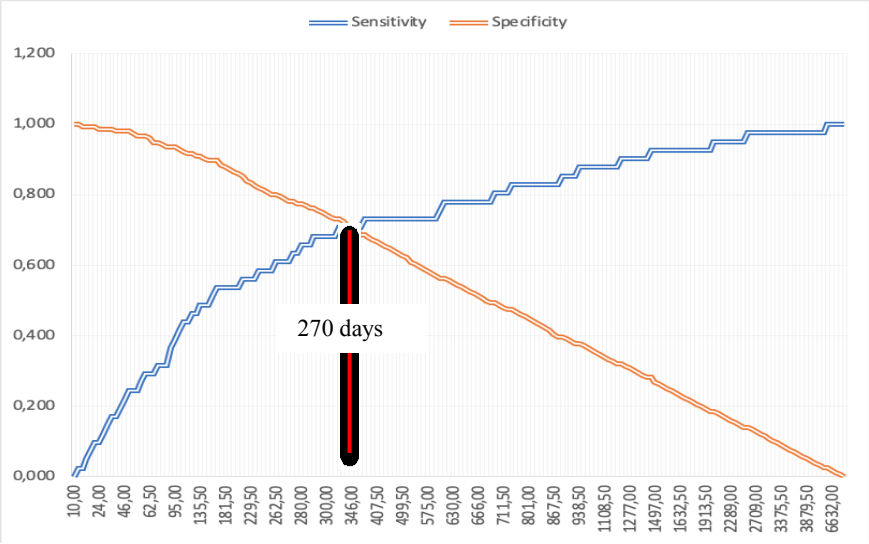
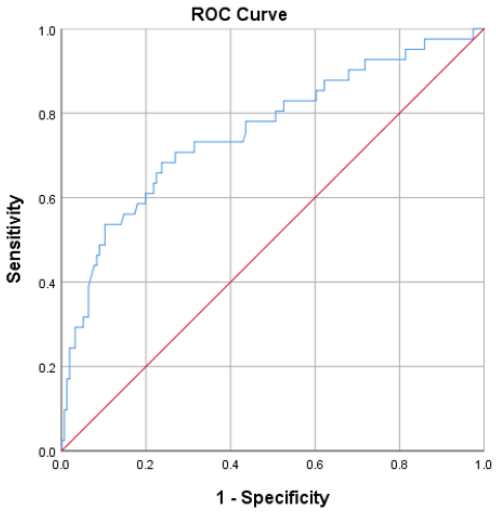


Figure 4

Age Cut-Off in Predicting Mortality



We evaluated six potential predictors of mortality after the BT shunt procedure among these subjects. Based on the bivariate analysis, four predictors showed significant association with mortality. They are age, weight <4.25 Kg, type of surgery (elective/emergency), and surgical approach. Weight <4.25 Kg and emergency surgery significantly affected the mortality after the BT shunt procedure. Weight under <4.25 Kg increased the risk of mortality 20 times, while emergency procedure raised the risk 3.5 times (Table 3).

Table 3.
Bivariate and Multivariate Analysis Of Mortality

Variables	Mortality		Bivariate		Multivariate	
	Yes (n = 41)	No (n = 156)	RR (95% CI)	*p*	OR (95% CI)	*p*
Age, n (%)						
≤270 days	25 (44,6)	31 (55,4)	3,934 (2,279-6,790)	<0,0001	2,418 (0,742-7,880)	0,143
>270 days	16 (11,3)	125 (88,7)				
Weight, n (%)						
<4,25 Kg	21 (70,0)	9 (30,0)	5,845 (3,641-9,382)	<0,0001	20,867 (7,372-59,07)	<0,0001
≥4,25 Kg	20 (12,0)	147 (88,0)				
Nutritional status, n (%)						
Malnutrition	22 (21,2)	82 (78,8)	1,035 (0,600-1,788)	1,000		
Good Nutritional Status	19 (20,4)	74 (79,5)				
Type of Surgery, n (%)						
Urgency/Emergency	13 (35,1)	24 (64,9)	2,008 (1,156-3,487)	0,031	3,463 (1,264-9,489)	0,016
Elective	28 (17,5)	132 (82,5)				
Surgical approach, n (%)						
Median sternotomy	32 (35,6)	58 (64,4)	4,227 (2,132-8,380)	<0,0001	1,588 (0,497-5,074)	0,435
Thoracotomy	9 (8,4)	98 (91,6)				
Postoperative transfusion, n (%)						
Yes	19 (23,2)	63 (76,9)	1,211 (0,703-2,088)	0,61		
No	22 (19,1)	93 (80,9)				

The mortality trend after the BT shunt procedure between 2016 – 2020 can be seen in Table 4, which shows a downward trend over the years. The total mortality from 2016 – 2020 was 41 (20,8%).

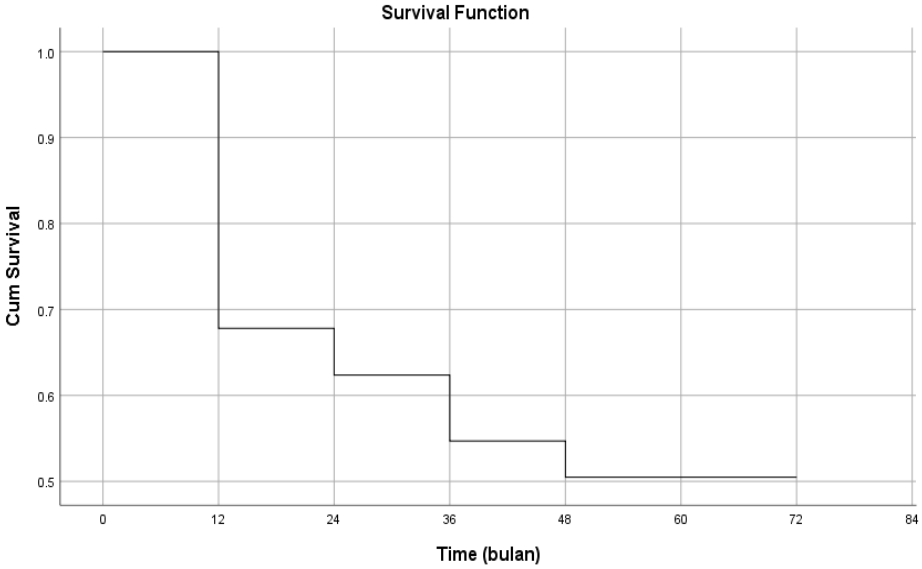
Table 4
Mortality of BT Shunt per Year

Mortality rate per year	n (%)
2016	12/38 (31,6)
2017	7/42 (16,7)
2018	12/47 (25,5)
2019	8/45 (17,8)
2020	2/25 (8)

The Kaplan Meier curve shows that the survival rate post BT Shunt also shows a downtrend over the years. The first year post operation shows a survival rate of 68%, followed by a survival rate of 62% in the second year, 55% in the third year, and a survival rate of 50% in the fourth and the fifth year post BT Shunt operation (Figure 5).

Figure 5

Kaplan Meier Curve of 5 Years Survival Post BT Shunt Operation



Discussion

The median of patients' weight and age was 7.9 kg and 20 months when the procedure was performed. In this study, many older children underwent surgery due to the complex anatomy of defects and sub-development of pulmonary artery branches. This finding was in contrast with data from developing countries that mostly came from younger children. The previous BT shunt research at our center also had a wide range of ages, from 15 days to 17 years old.[7] A study in Canada had a median age of 8 days, and the mean weight was 3.1 Kg.[3] This apparent difference might be caused by the gap in healthcare access that remains a significant challenge in developing countries. Most patients (47.2%) had good nutritional status. However, 24.4% of patients had mild-moderate malnutrition, while the other 15.7% had severe malnutrition. Malnutrition is a common finding in CHD. The incidence of malnutrition-related CHD in Indonesia is 70.7%, and 22.4% is severe.[8] Previous studies also reported that children with CHD tend to have height and weight under the 50th percentile; therefore, failure to thrive is expected in this cohort.[9] Several factors are involved in this malnutrition tendency, such as inadequate intake, increase metabolic requirements, and the abnormality in absorption secondary to CHD.[9,10] Early intervention in CHD is required to improve children's nutritional status. A palliative procedure like BT shunt can help patients to improve their condition before the definitive correction surgery. However, it is vital to notice that the outcomes of BT shunt are also poorer in children with severe malnutrition.[9,10]

One of the most important predictors of palliative procedures outcomes is age. Cardiac surgery is ideally performed at a younger age to lower morbidity and mortality. However, not every child is eligible for surgery due to anatomical complexity, hemodynamic status, and pulmonary vascular resistance. It is vital to perform the palliative procedure at the optimal age as it can affect the outcomes significantly.[11] In this study, the patients' median age was 20 months when the procedure was performed.

The thoracotomy approach was utilized more often than sternotomy for the BT shunt procedure at our center (54.3% vs. 45.7%). This approach is considered more accessible, faster, and safer. However, a study in Boston reported that thoracotomy had a higher failure rate

compared to sternotomy.[12] One of the common complications is phrenic nerve paralysis, with an incidence of 23.8% in patients underwent this procedure via thoracotomy.[13,14]

However, only two predictors, weight <4.25 Kg and type of surgery (elective/emergency), significantly affected the mortality after the BT shunt procedure. Weight under <4.25 increased the mortality 20.8 times, while the emergency type of surgery raised the mortality chance 3.5 times. No study describes the association between type of surgery (elective/emergency) with mortality risk.[6,15] The reason why low bodyweight may predispose patients to death was the unavailability of grafts with a size under 3 mm.

Finally, this study helps describe the outcomes after BT shunt in Indonesia, which the data were rarely available previously. The study's population was also large enough to describe these outcomes. A multicenter approach might also be important to be adopted in the future because of the high variability in experience and facilities between centers in Indonesia.

Conclusion

The mortality rate after BT shunt at PJT Rumah Sakit Cipto Mangunkusumo was 20.8%. Based on multivariate analysis, the predictor factors associated with mortality were weight <4.25 kg and emergency procedures.

Patient Consent Form: All participants were informed about subject of the study.

Author's Contribution: A.P. and E.M. gave substantial contributions to the conception or design of the work in acquisition, analysis, or interpretation of data for the work. A.P, A.K., and E.M. had a part in article preparing for drafting or revising it critically for important intellectual content. A.P, A.K., and E.M. gave final approval of the version to be published and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Conflicts of interest: There are no conflicts of interest.

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