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ARTICLE

Growth of the Pulmonary Valve Annulus after the Modified Blalock-Taussig Shunt in Patients with Tetralogy of Fallot

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ABSTRACT

Background: The surgical outcomes of tetralogy of Fallot (TOF) have evolved dramatically and have resulted in lower mortality rate. Currently, the many cardiac centers have a trend to early single-stage complete repair more than a staged repair. However, the patients who have an early primary repair were required transannular patch augmentation of a pulmonary valve frequently. This effect has been developed a chronic pulmonary insufficiency may lead to right ventricular dilation, dysfunction. In this era, the aim of treatment of TOF is attempted to preserve pulmonary valve annulus for prevent right ventricular dysfunction in the future. The systemic to pulmonary artery shunt is a palliative procedure or known as staged repair for symptomatic patients with TOF. The modified Blalock-Taussig shunt (mBTS) is the most useful systemic to pulmonary shunt and perform as an initial procedure before complete repair. The mBTS can provide increase pulmonary blood flow as well as improve oxygenation and also promote pulmonary artery (PA) growth. However, the effect of this procedure to promote growth of a pulmonary valve annulus is still debate. **Objectives:** To compare a growth of pulmonary valve annulus between after staged repair and primary repair in patients with TOF (without pulmonary atresia). **Methods:** A retrospective case-control study, review of patients with TOF underwent total repair at our hospitals from January 2005 and December 2017 was performed, a total number of 112 patients underwent TOF repair. Twenty-nine patients (26%) underwent a staged repair (mBTS group) and 83 (74%) underwent total repair only or primary repair (PR group). We evaluated diameter of pulmonary valve annulus by using echocardiography at the time of first diagnosis and before complete repair on both groups. **Results:** The age of diagnosis of mBTS group were younger than PR group (p = 0.011). Therefore, pulmonary valve annuls were smaller in mBTS group. (Z-score, −2.93 ± 1.42 *vs.* −1.89 ± 0.97; p = 0.001). However, the growth potential of pulmonary valve annulus was increase more than PR group significantly (Z-score, −1.46 ± 1.02 *vs.* −2.11 ± 1.19; p = 0.009) Even though a patent ductus arteriosus was found commonly in PR group (p = 0.018). **Conclusions:** Our results suggest the systemic to pulmonary shunt or mBTS can promote growth of pulmonary valve annulus in patients with TOF.

KEYWORDS

Tetralogy of Fallot; systemic to pulmonary shunt; modified Blalock-Taussig shunt; pulmonary valve annulus; pulmonic valve Z score

1 Introduction

Tetralogy of Fallot (TOF) is the most common congenital cyanotic heart disease, characterized by large ventricular defect (VSD), right ventricular hypertrophy, right ventricular outflow tract (RVOT) obstruction, and over-riding of the aorta [1]. Since the first surgical palliative treatment for TOF was performed by Alfred Blalock at Johns Hopkins University in 1945 [2,3]. The first complete repair was successful by Lillehei at the University of Minnesota in 1954 [4,5]. In 1975, de Leval invented a modified technique, by using a polytetrafluoroethylene interposition graft known as the modified Blalock-Taussig shunt (mBTS) [6]. The options for treatment of symptomatic neonates and young infants with TOF include both early primary repair and staged repair with mBTS. The early primary repair of TOF has the advantage of achieving a normal circulation earlier, less right ventricle hypertrophy, single operation and financial benefits [7]. However, the neonate or young infant who had an early primary repair were required transannular patch augmentation more than staged repair [8,9]. The result of transannular patch has been developed a chronic pulmonary insufficiency lead to right ventricular dilation and dysfunction in a future [10–12].

Recently, the Society of Thoracic Surgeons database present the mortality rate of mBTS is higher than a primary repair in symptomatic patients [13]. Nowadays, the surgical options for treatment patients with TOF was transition from staged repair to primary repair. Some cardiac centers consider an early primary repair in asymptomatic neonate to be face with many postoperative problems and hence, still prefer mBTS to palliate in neonatal period [14–16] and also these problems were occurred in symptomatic neonate with TOF.

Nevertheless, the optimal surgical strategy for symptomatic neonates and infants with TOF are still debate between with primary repair and staged repair. The effect of initial palliation with a systemic to pulmonary shunt regarding to growth of the pulmonary arteries had been published but resulted in expansion of pulmonary valve annulus still unclear [17]. This reason led to our research to evaluate a systemic to pulmonary shunt can be promote growth of pulmonary valve annulus.

2 Material and Methods

2.1 Ethical Approval

This study protocol was approved by the Institutional Review Board (IRB) of the Faculty of Medicine, Naresuan University (IRB No. 0851/61, Approval No. 690/2018) following the principles of the Declaration of Helsinki and its contemporary amendment. The Naresuan University Institutional Review Board approved a waiver of documentation of informed consent.

2.2 Study Population

A retrospective review of all consecutive children undergoing tetralogy of Fallot repair between January 2005 and December 2017 at Naresuan University Hospital and Buddachinaraj Hospital were performed. All patients are located at the lower northern of Thailand.

The objective was determined a growth of pulmonary valve annulus after mBTS or staged repair (mBTS group) to compare with primary repair (PR group) or single stage complete repair.

A total number of 112 patients underwent a complete surgical repair from the lower northern region of Thailand. Twenty-nine patients (26%) underwent a staged repair (mBTS group) and 83 (74%) underwent a primary repair (PR group). The preoperative demographics and clinical characteristics are show in Tab. 1.

The data include gender, age, body weight and oxygen saturation were record. The pulmonary valve annulus, main pulmonary artery (MPA), right pulmonary artery (RPA), left pulmonary artery (LPA) and McGoon ratio by using transthoracic echocardiography at the first time of diagnosis (before mBTS) and before complete surgical repair on both groups. The operative data were included timing of operation, pulmonary valve morphology. All patients received routine follow-up by a pediatric cardiologist. The mean duration of follow-up was 7.96 ± 4.28 years of both groups.

Table 1: The preoperative demographics and clinical characteristics at the first time of diagnosis

Variable	Total patients (n = 112)	Primary repair (n = 83)	Stage Repair (n = 29)	*P*-value
Gender (male)	66 (58.9%)	50 (60.2%)	16 (55.2%)	0.633
Age (days)	214.34 ± 344.29	247.83 ± 386.29	118.48 ± 141.62	0.011*
BW (kg)	5.97 ± 2.22	6.3 ± 2.29	5.04 ± 1.73	0.008*
Height (cm)	63.51 ± 10.03	64.97 ± 10.43	59.34 ± 7.48	0.009*
Preterm	6 (5.4%)	4 (4.8%)	2 (6.9%)	0.669
Low birth weight	11 (9.8%)	6 (7.2%)	5 (17.2%)	0.119
Oxygen saturation (%)	81.4 ± 7.79	84.63 ± 5.45	72.17 ± 5.89	<0.001*
DiGeorge syndrome	3 (2.7%)	3 (3.6%)	0 (0%)	0.299
Down syndrome	1 (0.9%)	1 (1.2%)	0 (0%)	0.553
VSD Type (%)				
Perimembranous	103 (92%)	75 (90.4%)	28 (96.6%)	0.291
Outlet VSD	9 (8%)	8 (9.6%)	1 (3.4%)	
Add mVSD	3 (2.7%)	2 (2.4%)	1 (3.4%)	0.766
Bilateral SVC	7 (6.3%)	7 (8.4%)	0 (0%)	0.106
PDA	14 (12.5%)	14 (16.9%)	0 (0%)	0.018*
Right side arch	38 (33.9%)	28 (33.7%)	10 (34.5%)	0.942
PV morphology				
Unicuspid	10 (8.9%)	7 (8.4%)	3 (10.3%)	0.756
Bicuspid	80 (71.4%)	56 (67.5%)	24 (82.8%)	0.117
Tricuspid	22 (19.6%)	20 (24.1%)	2 (6.9%)	0.045*

Note: *$P < 0.05$; BW, body weight; VSD, ventricular septal defect; Add-mVSD, additional muscular VSD; SVC, superior venacava; PDA, patent ductus arteriosus; PV, pulmonary valve.

2.3 Surgical Intervention and Observation Indicators

Indications for stage palliation were frequent hypoxic spelling or profound desaturation (oxygen saturation less than 70%), or the presence small pulmonary arteries with McGoon ratio less than one and a half. If patients had mild symptoms, we tried to postpone complete repair until body weight more than 10 kg. We performed a mBTS via thoracotomy in all cases. This is a previous protocol for treatment patients with TOF. Nowadays, we try to total repair earlier for prevent hypoxic spell events and this is a reason why I enroll patients until December 2017. The role of management for primary repair (PR group). Almost cases were less symptom and present were present only mild cyanosis. The primary repair was reserved for body weight near 10 kg and the anatomy of pulmonary arteries were suitable for

complete repair (McGoon ratio > 1.5). The exclusion criteria were patients with TOF with canal defect, pulmonary atresia, absent pulmonic valve, isolate single pulmonary artery or associated with other complex congenital heart disease.

2.4 Statistical Analysis

Statistical analysis was carried out with SPSS version 16.0 software (SPSS, Inc., Chicago, IL, USA). Continuous variables are expressed as mean standard deviation. Nominal variables were comparisons between the 2 groups were analyzed using the Mann-Whitney U-test and the chi-square test. The *P*-value < 0.05 was regarded as statistically significant.

3 Results

3.1 The Demographic Data and Patient Characteristics at the First Time of Diagnosis

The basic patient characteristics are presented in Tab. 1 as above. No significant difference was found in gender or prematurity or low birth weight or chromosomal abnormality (DiGeorge and Down syndrome) between both groups. However, the patient in mBTS group were younger than PR group (p = 0.011). Additionally, a body weight (P = 0.008), height (P = 0.011) and oxygen saturation (P = < 0.001) were lower than to PR group significantly.

3.2 The Echocardiographic Data at the First Time of Diagnosis

The echocardiography at the first time of diagnosis was demonstrated in Tab. 2. The echocardiographic data were correlate with patient characteristics that all right-side cardiac structure in mBTS group smaller than PR group significantly.

Table 2: Initial echocardiography data at the first time of diagnosis

Variable	Total patients (n = 112)	Primary repair (n = 83)	Stage Repair (n = 29)	Mean difference (95%CI)	*P*-value
PV (mm)	6.83 ± 1.67	7.19 ± 1.57	5.8 ± 1.55	1.39 (0.72, 2.06)	<0.001*
PV (Z-score)	−2.16 ± 1.19	−1.89 ± 0.97	−2.93 ± 1.42	1.05 (0.47, 1.62)	0.001*
mPA (mm)	6.65 ± 1.78	7.12 ± 1.63	5.31 ± 1.51	1.81 (1.12, 2.49)	<0.001*
mPA (Z-score)	−2.61 ± 1.27	−2.21 ± 0.97	−3.73 ± 1.36	1.52 (0.96, 2.07)	<0.001*
RPA (mm)	5.66 ± 1.45	6.02 ± 1.3	4.65 ± 1.38	1.36 (0.8, 1.93)	<0.001*
RPA (Z-score)	−0.37 ± 1.14	−0.08 ± 0.98	−1.21 ± 1.16	1.14 (0.7, 1.58)	<0.001*
LPA (mm)	5.65 ± 1.53	5.98 ± 1.43	4.69 ± 1.44	1.3 (0.69, 1.91)	<0.001*
LPA (Z-score)	0.17 ± 1.21	0.45 ± 1.06	−0.64 ± 1.26	1.09 (0.61, 1.57)	<0.001*
McGoon ratio	1.63 ± 0.19	1.69 ± 0.17	1.46 ± 0.12	0.22 (0.16, 0.29)	<0.001*

Note: *P < 0.05; PV, pulmonary valve; mPA, main pulmonary artery; RPA, right pulmonary artery; LPA, left pulmonary artery.

3.3 The Demographic Data of Modified Blalock-Taussig Shunt

In total, 29 (26%) patients had palliative procedures before complete repair. The median age at mBTS was 2.6 months and the median body weight was 5 kg. The detail of the palliative procedure is summarized in Tab. 3. In our series, all patients were survived after mBTS. Complications after mBTS included shunt stenosis 6 (20.7%) patients that were need intervention. A mBTS was induce the pulmonary arteries branch stenosis 8 (25.5%) cases.

Table 3: The demographic data of modified Blalock-Taussig shunt

Data (total = 29 patients)	Statistic
Age at time of mBTS (month)	2.73 ± 18.62
BW at time of mBTS (Kg)	5.75 ± 1.57
Height at time of mBTS (cm)	62.41 ± 7.37
Surgical approach mBTS	
Right mBTS	25 (86.2%)
Left mBTS	4 (13.8%)
Central mBTS	0 (0%)
Diameter of graft (mm)	4.43 ± 0.53
Complication mBTS stenosis	
No	23 (79.3%)
yes	6 (20.7%)
Cardiac intervention after mBTS	
No intervention	22 (78.6%)
Balloon dilate	3 (10.7%)
Stent mBTS	3 (10.7%)
mBTS induce PA branches stenosis	
no	21 (72.5%)
RPA stenosis	7 (24.1%)
LPA stenosis	1 (3.4%)
Both PAs stenosis	0 (0%)
Dead or survival after MBTS	
Death	0 (0%)
Survive	29 (100%)

3.4 The Echocardiographic Data at the Time before Complete Repair

At the time of total repair are depicted in Tab. 4. There were no significant differences in age, weight and height between of both groups. However, the pulmonary valve annulus of mBTS group was increased in size from −2.93 (−1.51 to −3.72) to −1.46 (−2.48 to −0.44) in z-score ($P = < 0.001$) and also larger than the PR group ($P = 0.009$) significantly at the time of total repair. The pulmonary valve annulus z-score in the PR group was tendency to reduce when to be compare with initial diagnosis ($P = 0.058$). It is displayed in Fig. 1.

The detail of all patients in each case on both groups were demonstrated in Fig. 2. The mean duration of follow-up was 7.96 ± 4.28 years (mBTS group, 6.75 ± 3.57 years; PR group, 8.36 ± 4.44 years; $P = 0.090$), there were not significantly different between both groups.

Table 4: The echocardiographic data at the time before complete repair

Variable	Total patients (n = 112)	Primary repair (n = 83)	Stage repair (n = 29)	Mean difference (95%CI)	*P*-value
Age (days)	1062.75 ± 631.44	1036.87 ± 600.93	1136.83 ± 717.93	-99.96 (-370.46, 170.54)	0.466
BW (Kg)	12.01 ± 3.66	11.96 ± 3.87	12.17 ± 3.03	-0.21(-1.78, 1.36)	0.794
Height (cm)	90.12 ± 12.87	89.29 ± 13.12	92.52 ± 12	-3.23 (-8.72, 2.26)	0.246
PV (mm)	9.65 ± 2.64	9.35 ± 2.53	10.51 ± 2.8	-1.16 (-2.27, -0.04)	0.042*
PV (Z-score)	−1.94 ± 1.18	−2.11 ± 1.19	−1.46 ± 1.02	-0.65 (-1.15, -0.16)	0.009*
mPA (mm)	8.55 ± 3.31	8.44 ± 3.11	8.84 ± 3.86	-0.4 (-1.82, 1.02)	0.579
mPA (Z-score)	−2.43 ± 1.23	−2.51 ± 1.26	−2.19 ± 1.12	-0.32 (-0.85, 0.2)	0.223
RPA (mm)	8.8 ± 2.15	8.56 ± 1.71	9.5 ± 3	-0.94 (-2.14, 0.25)	0.118
RPA (Z-score)	0.41 ± 1.04	0.22 ± 0.9	0.94 ± 1.24	-0.72 (-1.22, -0.21)	0.006*
LPA (mm)	8.72 ± 2.19	8.65 ± 1.99	8.9 ± 2.72	-0.25 (-1.19, 0.69)	0.593
LPA (Z-score)	1.08 ± 0.99	1.01 ± 0.97	1.27 ± 1.02	-0.27 (-0.69, 0.16)	0.215
McGoon ratio	1.94 ± 0.22	1.9 ± 0.23	2.03 ± 0.17	-0.13 (-0.23, -0.04)	0.005*

Note: *$P < 0.05$; PV, pulmonary valve; mPA, main pulmonary artery; RPA, right pulmonary artery; LPA, left pulmonary artery.

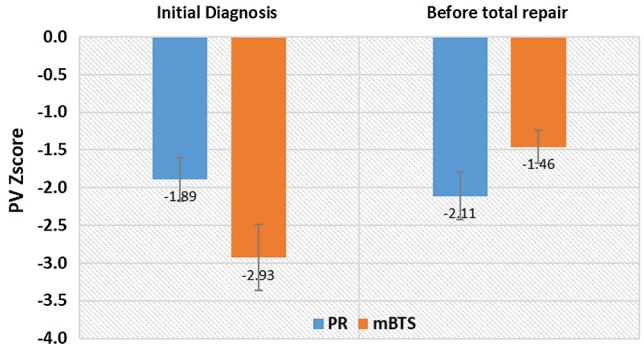

Figure 1: The pulmonary annular valve (Z-score) growth to compare between PR and mBTS group

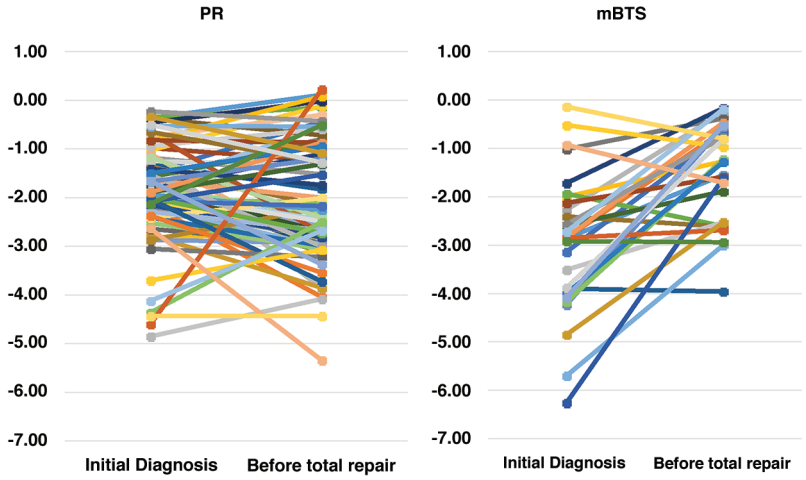

Figure 2: Line chart of the pulmonary valve annular z-score growth of both groups

4 Discussions

In this era, the trend of surgical treatment for TOF is toward to early primary repair. However, many centers were concerned about this technique need a transannular patch for enlargement of right ventricular outflow tract frequently and also increased postoperative morbidity when operated during early infant [18–20]. In our unit, we have been a policy to early primary repair in early infant with TOF. Whenever we performed an early primary repair, may need to do transannular patch. Which is against to the principle for treatment TOF that attempt to preserve pulmonary valve annulus. Currently study from Japan [21], present the theory the mBTS can promote growth of pulmonary valve annulus. This article explained about the effect of a shunt can increase pulmonary blood flow then raised blood returned to the left side heart and also increased blood flow pass through VSD. This effect can increase preferential blood flow pass through pulmonary valve annulus that demonstrated by a flow visualization method under echocardiography. However, this hypothesis is quite unclear and need further study to support.

In our study, we tried to compare a potential growth of pulmonary valve annulus between a staged repair (mBTS effect) and patients underwent primary repair (non-shunt effect or natural growth). Our result demonstrated the pulmonary valve annulus has a potential growth after mBTS. In our study, we perform a mBTS via thoracotomy in all cases there is a different from previous reports that operated via a median sternotomy [21–23]. Usually, when we did mBTS via a thoracotomy, we need a length of graft longer than a median sternotomy approach. The length of graft may cause to increase size of pulmonic valve annulus should be concerned. However, our study demonstrated a mBTS via thoracotomy can promote growth of pulmonary valve annulus when you used graft longer than median sternotomy approach. Our results appeared to be compatible with the other studies [21–23]. Therefore, the different about length of graft not effected to growth of pulmonic valve annulus. The advantage of thoracotomy approach, we can easily to perform complete repair which were not interfere by previous surgery. In addition, the mBTS can promote growth of RPA and also increase McGoon ratio. Anyway, the LPA and MPA diameter was not different between both groups because most case (86.2%) were performed right mBTS.

Furthermore, the true benefit of mBTS is not consider only the pulmonary valve growth but should determine this procedure able to increase rate of pulmonary valve sparing during a complete surgical repair.

In our study had restriction inherent in retrospective observational data studies. We suggest have a further investigation about rate of pulmonary valve preservation and can be delay pulmonary regurgitation or more preserve right ventricular function after TOF repair.

5 Conclusions

Our study support a modified Blalock-Taussig shunt is a valuable procedure for promote pulmonary valve annular growth in patients with TOF.

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