

Mid and Long Term Evaluation of Left MBTS after Cavopulmonary Shunts: A Retrospective Cohort Study

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Abstract

Background: Congenital heart diseases are the most common type of congenital malformations. Some of them may pass unnoticeable in the first days or maybe years of life, but the others are pretty serious and life-threatening due to major malformations that lead to the mixing of oxygenated and deoxygenated blood. Most of these cases are not repairable and require separating the venous and arterial blood to help improve oxygen saturation.

Aim of Study: The aim of the study is to evaluate the mid and long-term effects of left MBT shunt after bidirectional cavopulmonary shunt.

Patients and Methods: A retrospective cohort study was conducted on 20 patients who underwent mBTS after BDG. We evaluated the mid and long-term effects of mBTS in those patients especially SpO₂ and echocardiographic findings. The study considered the ethical principles of the Helsinki Declaration approval from the research ethics committee at our institution (MS 510/2023).

Results: There was a significant increase in postoperative SpO₂ in comparison to preoperative SpO₂ ($p < 0.001$) with a mean difference of 20.95%.

Conclusion: Congenital cyanotic heart diseases represent a life-threatening condition where the main problem is desaturation due to the mixing of arterial and venous blood. BDG is a step to separate the oxygenated and deoxygenated blood. But later on, oxygen saturation continues to decline and further intervention is needed. In this study, we found that mBTS provides a post-operative significant increase in oxygen saturation in a wide group of those patients even who are not candidates for other operations without significant complications.

Key Words: Congenital heart diseases – MBTS – Cavopulmonary Shunts.

Introduction

CONGENITAL heart diseases are the most common type of congenital malformations [1]. Some

of them may pass unnoticeable in the first days or maybe years of life, but the others are pretty serious and life-threatening due to major malformations that lead to the mixing of oxygenated and deoxygenated blood. Most of these cases are not repairable and require separating the venous and arterial blood to help improve oxygen saturation [2].

As the main problem here is desaturation due to the mixing of arterial and venous blood, trials were made to create another pathway to help improve oxygen saturation and separate the arterial and venous blood. Cavopulmonary shunts were described as a possible solution for long-standing cyanosis [3].

Bidirectional Glenn is one of the cavopulmonary shunts that aims at improving the oxygen saturation where the SVC is transected at its attachment with the right atrium and anastomosed to the pulmonary artery [4]. Although BDG relieves the cyanosis for a period of time, most cases develop desaturation with advanced age and further intervention is needed [5].

Fontan operation is performed as a second stage of repair where the IVC with its junction with the right atrium is connected to the pulmonary artery. This procedure results in complete separation of pulmonary and venous return (total cavo-pulmonary shunt) [6] but it has many restrictions and many complications, especially in the long term. Most of the complications are caused by chronic systemic venous hypertension due to the lack of a pumping sub-pulmonary ventricle. The most frequent complications are arrhythmias, thromboembolism, protein-losing enteropathy, heart failure and plastic bronchitis [7]. The pathogenetic mechanisms are not completely understood but they are all connected to chronic systemic venous hypertension. Quality of life and life expectancy are deeply impaired by the onset of such complications [8].

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While searching for options for dealing with the effects of such complex congenital heart problems—mainly desaturation it was observed that patients with complex cardiac anomalies in addition to patent PDA have better blood oxygenation than those without patent PDA [9].

Depending on the fact that arterial shunts could improve oxygen saturation and regarding the restrictions and complications of the Fontan procedure, the idea of performing an arterial shunt like MBT shunt in patients with BDG suffering from cyanosis came up.

The Blalock-Taussig's shunt was initially introduced by Alfred Blalock and Vivien Thomas [10], who used the subclavian artery as an anastomosis to create a canine model of PA hypertension. This idea was then adopted as a last-ditch effort to increase pulmonary blood flow in tetralogy of Fallot patients [11].

Over time, a major improvement of the traditional Blalock-Taussig shunt was devised, avoiding the subclavian artery by using a synthetic vascular prosthesis. Compared to the original BT shunt, this modification had a number of benefits, including a decreased propensity to deform hypoplastic PAs, a reduced requirement for mediastinal dissection, the preservation of upper extremity blood flow, a steady shunt flow, and an appropriate length [12]. The modified Blalock Taussig shunt is still the most popular systemic PA shunt in use today because of these important benefits [13]. Additionally, the hepatic factors to the lungs are preserved by MBT shunt following BDG, which lowers the risk of AV malformation [14].

Aim of the study:

The aim of the study is to evaluate the mid and long-term effects of left MBT shunt after bidirectional cavopulmonary shunt.

Patients and Methods

Study setting:

This study is a retrospective cohort study from January 2023 to December 2023 on 20 patients was conducted at the Department of Cardiothoracic Surgery Ain Shams University Hospital.

Inclusion criteria:

Patients >5 years old, patients with cavopulmonary shunt operation, patients with sizable left pulmonary artery, patients with normal anatomy of the left subclavian artery, patients with oxygen saturation less than 75%.

Exclusion criteria:

Patients ≤5 years old, patients with dysfunctioning cavopulmonary shunt, patients with other debilitating diseases expect that may affect life expectancy.

Procedures and tools:

Regarding medical records of the patients:

Demographics (age, sex, BMI), complete history taking including past medical history of any other illness, general examination including: Vital signs, collection of pre-operative data: BMI, oxygen saturation, echocardiography.

Surgical technique of MBTS:

The patient is put in the left lateral position for the posterolateral thoracotomy to be done, dissection over the left subclavian artery and the main pulmonary artery and its left branch, vascular clamps are applied and Side to side anastomosis is done using a gortex tube graft of suitable size, removal of vascular clamps and checking for bleeding from the anastomosis, placement of chest tube and primary closure in layers.

Collection of post-operative data:

Oxygen saturation, NYHA class, echocardiography, post-operative morbidity and mortality.

Statistical analysis:

Statistical analysis was performed with the SPSS Version 20.0 statistic software package. Categorical data were expressed as number and percent. Continuous data were expressed as mean, standard deviation (Std. Deviation), median, minimum, and maximum. Comparisons of pre-operative and post-operative continuous data were performed using paired-samples *t*-test. A value of $p < 0.05$ was considered statistically significant.

Results

The results are discussed in detail in and will evaluate the outcome of mid and long-term effects of left MBT after bidirectional cavopulmonary shunt.

Table (1): Gender of the studied patients.

Gender	Number	Percent
Male	14	70
Female	6	30

The study included 20 patients underwent MBTS following CPS via thoracotomy approach. There were 14 males (70%) and 6 females (30%). (Table 1).

The indications of mBTS were hypoplastic left ventricle in 30%, TGA+ pulmonary atresia in 15%, D-TGA in 15%, TGA+DORV in 15%, DILV+PS in 10%, DORV+VSD in 5%, DORV in 5%, and TOF with hypoplastic left ventricle in 5%.

Preoperative left ventricular ejection fraction (LVEF) ranged between 52 and 65% with a median of 58.50% and a mean of 58.90±4.25%.

Table (2): Durations of postoperative ICU and hospital stays.

Statistics	ICU stay (days)	Hospital stay (days)
Mean	5.40	14.35
Median	5	13.50
Std. Deviation	1.23	4.15
Minimum	4	8
Maximum	8	22

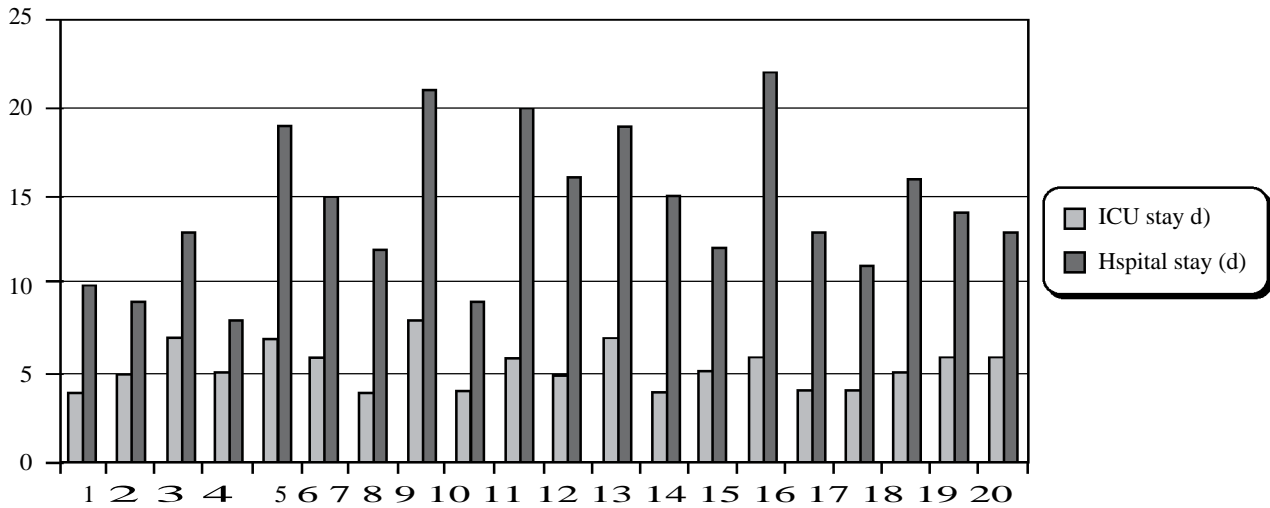


Fig. (1): Postoperative ICU and hospital stay by days for each patient.

ICU stay ranged between 4 and 8 days with a median of 5 days and a mean of 5.40±1.23 days. Postoperative hospital stay ranged between 8 and 22 days with a median of 13.50 days and a mean of 14.35±4.15 days. (Table 2) (Fig. 1).

Table (3): Postoperative oxygen saturation (SpO₂).

Statistics	Postoperative SpO ₂
Mean	88.90
Median	89
Std. Deviation	2.40
Minimum	85
Maximum	94

Postoperative oxygen saturation (SpO₂) ranged between 85 and 94% with a median of 89% and a mean of 88.9±2.40%. (Table 3).

Table (4): Change in postoperative oxygen saturation (SpO₂).

Statistics	Postoperative-Preoperative SpO ₂
Mean difference (%)	20.95
p-value	<0.001*

*Significant difference.

There was a significant increase in postoperative SpO₂ in comparison to preoperative SpO₂ ($p < 0.001$) with a mean difference of 20.95%. (Table 4).

The postoperative course was uneventful in the majority of cases. There was no mortality, re-operation, or shunt occlusion. There were minimal postoperative complications reversed on medical treatment in 3 cases (15%) including thrombosis treated by anticoagulants, wound infection, and pneumonia in one patient for each (5%).

Discussion

Structural anomalies of the heart or intrathoracic great vessels that happen during fetal development are known as congenital heart disorders (CHD). The most prevalent kind of birth abnormality and the main reason why children with congenital abnormalities die is congenital heart disease (CHD) [1]. There are two types of congenital heart disease (CHD): Cyanotic CHD, often known as critical congenital heart disease, and non-cyanotic CHD [15].

The main problem in cyanotic congenital heart diseases is desaturation due to the mixing of arterial and venous blood. Cavopulmonary shunts were described as a possible solution for long-standing cyanosis as they can provide another pathway to separate the arterial and venous blood [3].

Bidirectional Glenn is one of the cavopulmonary shunts that aims at improving the oxygen saturation where the SVC is transected at its attachment with the right atrium and anastomosed to the pulmonary artery [4]. Although BDG relieves the cyanosis for a period of time, most cases develop desaturation with advanced age and further intervention is needed [5].

Fontan operation is performed as a second stage of repair and it results in complete separation of pulmonary and venous return [6]. But it has many restrictions and complications, especially in the long term.

However, in the event of a subpulmonic pumping chamber's absence, the unphysiological Fontan circulation leads to a decrease in cardiac output and an increase in central venous pressure, which can lead to a number of short- or long-term problems.

These complications include: [18]

One common cardiac complication in Fontan circulation is atrioventricular valve regurgitation (AVVR), which has a negative effect on long-term results by raising the likelihood of mortality or heart transplantation [19].

Following Fontan surgery, arrhythmias such as bradyarrhythmia, supraventricular tachyarrhythmia, and atrial tachyarrhythmia are frequently seen. They might result in cardiac failure, thrombogenesis, AVVR worsening, or even abrupt death [20].

Ventricular dysfunction, either diastolic or systolic, is a frequent consequence that worsens long-term mortality and morbidity. There are several contributing factors, including as repeated operations, pressure and volume overloading, and persistent hypoxia [21].

In Fontan circulation FSV patients, thromboembolic events (TE) are a major cause of morbidity and death. Despite the lack of clarity around the genesis and etiology of TE, endothelial dysfunction, aberrant hemodynamics, and hypercoagulability are the three components of Virchow's triad that are implicated. Endothelial dysfunction specifically refers to the loss of prothrombotic products and vasoconstrictors, along with a rise in prothrombotic factors and antithrombin, Protein C, S, and antithrombin, and vasodilators such nitric oxide, which ultimately leads to a prothrombotic state [22].

With a remitting and relapse course, protein-losing enteropathy is an uncommon but devastating complication. It is characterized by massive protein loss into the digestive tract, which results in low levels of serum albumin and total protein, hyper-

coagulability, and immunodeficiency due to enteric loss of lymphocytes and immunoglobulins [23].

Like PLE, plastic bronchitis (PB) is a rare but potentially catastrophic condition that causes cohesive and obstructive bronchial casts to form and expectorate in Fontan patients [24].

Liver fibrosis, cirrhosis, and possibly cancer are among the congestive hepatopathies that make up fontan-associated liver disease (FALD) [25].

At rest, the kidneys' blood supply normally makes up 20-25% of the heart's output. Here, renal impairment, including acute kidney injury (AKI) and chronic kidney disease (CKD), may result from the Fontan hemodynamic change [26].

In the review conducted by Johanna et al., Many patients with Fontan circulation experienced serious long-term complications such as difficulty in areas of cognition related to attention and executive functioning, visual-spatial reasoning, and psychosocial development. They were also at high risk for mental health morbidities, particularly anxiety disorders and depression [27].

It was an interesting remark that patients with patent PDA who did not undergo PDA closure during the cavopulmonary shunt operation had more acceptable saturation than patients with closed PDA, and they did not need further intervention [9]. Depending on the fact that arterial and pulsatile blood supply to the lung can help to avoid severe progressive desaturation, hence came the idea of doing arterial shunts after BDG.

Not all patients who have had a Glenn shunt are candidates for Fontan, in addition to the serious complications already mentioned. This is because many of these patients have significant A-V valve regurgitation, systemic ventricular dysfunction, pulmonary artery hypoplasia, and high pulmonary vascular resistance [17].

Therefore, many approaches were being tested to help individuals with BDG who experienced increasing desaturation increase their oxygen saturation. Venovenous collaterals have been reported to worsen the prognosis in these individuals by lowering their arterial oxygen saturation. Transcatheter closure for those collaterals was carried out in the Doaa et al., trial in an effort to raise the oxygen saturation [28].

Creating radial and axillary arteriovenous (AV) fistulae to augment pulmonary blood flow across the preexisting cavopulmonary shunt was another

strategy to aid in increasing oxygen saturation following BDG [29].

According to a study by William et al., a 13-year-old patient with tricuspid atresia who had a superior vena cava-right pulmonary artery (SVCRPA) shunt at age 3 could benefit from an A-V fistula as a useful way to supplement blood flow to the right lung. This was achieved by increasing the flow through the shunt, giving it a pulsatile character, and ultimately lowering the blood's viscosity [30].

Patients with complicated cardiac abnormalities who have a patent PDA tend to have greater oxygen saturation than those who do not. Furthermore based on earlier research mentioning the benefit of A-V fistulae in raising oxygen saturation in patients who had previously had cavopulmonary shunts. The notion of executing mBTS subsequent to the BDG was raised.

Previously referred to as the standard or classic Blalock-Taussig shunt (BT shunt), the modified Blalock-Taussig-Thomas shunt (mBTT shunt) is a palliative surgical operation used to treat patients with cyanotic cardiac disorders defined by reduced pulmonary artery flow [31].

The goal of the BT shunt is to deliver enough blood flow to the pulmonary artery to alleviate cyanosis without causing pulmonary overcirculation. Through a lateral thoracotomy, the subclavian artery was divided and anastomosed end-to-side to the pulmonary artery in the standard BT shunt surgery. After numerous modifications, the initial procedure became the mBTT shunt, which creates a systemic-pulmonary shunt without compromising any of the brachiocephalic tributaries or the subclavian arteries by using an interposition polytetrafluoroethylene (PTFE) graft [31].

The benefits of pulmonary artery shunts were listed in the review paper written by Pankaj et al., as follows: [32]

Enhancement of hematological parameters and systemic oxygen saturation: The creation of an axillary arterio-venous fistula for the purpose of correcting hypoxemia after a first BDG was documented by Glenn and Fenn in 1972 [30]. After a BDG, patients who have a systemic to pulmonary artery shunt have higher systemic oxygen saturation due to increased pulmonary blood flow, increased recruitment of the pulmonary capillary bed, and decreased pulmonary vascular resistance. This leads to a reduction in the risk of cerebral and peripheral venous thrombosis, a progressive decrease in hematocrit, blood viscosity, and coagulopathy, and an increase in oxygen saturation from 8% to 25% [33].

Increase in pulmonary artery size: Ishikawa et al. [34] have demonstrated significant increases in pulmonary artery size on the ipsilateral side following a Blalock-Taussig shunt, especially if the shunt was performed within the first year of life, despite conflicting data regarding the increase in pulmonary artery size following the creation of a systemic pulmonary artery shunt. It's unclear why many patients who have had a systemic to pulmonary shunt do not see a significant increase in the size of their pulmonary arteries after the procedure, but it's plausible that a relatively small shunt is not able to pump enough blood into the pulmonary circulation to match the normal cardiac output. An innate flaw in the pulmonary arteries or the integration of ductal tissue inside the pulmonary artery are two other potential causes.

Regression of pulmonary arteriovenous collaterals (malformations; AVM): The majority of patients experience pulmonary AVM 5 to 10 years after BDG; this results in a worsening of pulmonary function and an increase in cyanosis. The exact cause of these AVMs is unknown, but it is thought to be caused by a lack of hepatic factor circulating through the lungs after antegrade pulmonary blood flow is interrupted after a BDG [35]. According to a study by Doff B. et al., these AVMs regress and are prevented from growing further [16].

Improvement in ventricular function: Six patients who had undergone a BDG before and were deemed inappropriate for TCPC because of ventricular dysfunction were palliated using a brachial arteriovenous fistula in the Quarti et al., research [36]. After a brachial arteriovenous fistula was created, all six patients' ventricular function improved during follow-up, and they all underwent successful TCPC procedures within six years. It is unclear how precisely the systemic to pulmonary artery shunt in these individuals improved ventricular function. Ventricular function may have improved, nevertheless, because of factors such as decreased blood viscosity from a lower hematocrit, increased myocardial preload, and increased oxygen saturation from better oxygen delivery to the myocardium [36].

Out of 320 children with cyanotic congenital heart malformations who had previously undergone cavopulmonary shunt operations, 11 were deemed unsuitable for definitive repair, a Fontan procedure, or other palliation due to their increasing cyanosis and exercise intolerance, as reported by A Magee et al. [16]. Three individuals had a prior bidirectional cavopulmonary connection, and eight had a prior Glenn shunt. Ten patients underwent ipsilateral axillary arteriovenous fistula formation to increase pulmonary blood flow. Before surgery, mean

oxygen saturations were $80\% \pm 2\%$, immediately after surgery, they were $85\% \pm 2\%$, and after a mean follow-up interval of 7.4 years (range 0.1 to 15.5 years), they were $84\% \pm 3\%$. This suggested that when alternative choices are limited, establishing an axillary arteriovenous fistula to increase pulmonary blood flow following a cavopulmonary shunt offers helpful palliation for complicated cyanotic heart disease.

In this study, we reported the mid- and long-term postoperative outcomes, particularly oxygen saturation, after we retrospectively evaluated the data of 20 patients who underwent BDG in our hospital and received mBTS.

The preoperative oxygen saturation (SpO_2) had a mean of $67.95 \pm 3.56\%$ with a range of 60 to 73%. The median was 68.50%. The range of postoperative oxygen saturation (SpO_2) was 85-94%, with a mean of $88.9 \pm 2.40\%$ and a median of 89%. With a mean difference of 20.95%, there was a substantial rise in postoperative SpO_2 compared to preoperative SpO_2 ($p < 0.001$). Therefore, it was statistically significant that in patients who have had a prior cavopulmonary shunt and are not candidates for other procedures or cannot tolerate their consequences, left mBTS effectively raises oxygen saturation.

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Worldwide, a midline sternotomy has become the standard procedure for managing mBTS. It's also said that doing a mBTS via thoracotomy is technically more difficult. On the other hand, thoracotomy surgery has been suggested to put patients at lower risk for wound infections, postoperative respiratory impairment, shorter hospital stays, and cosmetic issues.

The sole side effect reported in the A Magee et al., research [16] was a patient's slight arm edema distal to the fistula; this patient's distal vein was left unligated and she needed another procedure after 19 days.

Most of the subjects in our dataset had unremarkable postoperative courses. No shunt obstruction, death, or re-operation occurred. Three cases (15%) had modest postoperative problems that were resolved with medical treatment; they included one patient's (5%), wound infection, and pneumonia, as well as thrombosis treated with anticoagulants.

Conclusion:

Congenital cyanotic heart diseases represent a life-threatening condition where the main problem is desaturation due to the mixing of arterial and venous blood. BDG is a step to separate the oxygenated and deoxygenated blood. But later on, oxygen saturation continues to decline and further intervention is needed. In this study, we found that mBTS provides a post-operative significant increase in oxygen saturation in a wide group of those patients even who are not candidates for other operations without significant complications.

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**تقييم الآثار متوسطة وبعيدة المدى لتحويله بلالوك-توسيج اليسرى
بعد إجراء تحويله أجوفية رئوية:
دراسة حشدية رجعية.**

المقدمة: فى رحلة البحث عن الخيارات المتاحة للتعامل مع أثر العيوب الخلقية المعقدة فى القلب - خاصة نقص الأكسجين - تم ملاحظة أن المرضى أصحاب العيوب الخلفية المعقدة بالقلب الذين لديهم القناة الشريانية السالكة يتمتعون بمستويات أفضل من أكسجين الدم أكثر من نظائرهم الذين لا يمتلكون القناة الشريانية السالكة.

هدف الدراسة: تقييم الآثار متوسطة وبعيدة المدى لتحويله بلالوك-توسيج اليسرى بعد إجراء تحويله أجوفية رئوية.

المرضى وطرق العلاج: تم إجراء دراسة حشدية رجعية تجمع المرضى الذين أجروا تحويله بلالوك توسيج اليسرى بعد جراحة جلين ثنائية الاتجاه. وتم أخذ وتم تقييم الآثار متوسطة وبعيدة المدى فى هؤلاء المرضى خاصة نسبة أكسجين الدم ونتائج فحوصات الأشعة التليفزيونية على القلب.

النتائج: تبين لنا من خلال جمع البيانات لعشرين مريضاً ممن أجروا تحويله بلالوك توسيج اليسرى بعد تحويله جلين ثنائية الاتجاه وجود فرق ذو دلالة إحصائية فى نسب أكسجين الدم للمرضى قبل وبعد تحويله بلالوك توسيج اليسرى. وجاءت نسبة المضاعفات لتمثل ١٥٪ من الحالات ولم يتم تسجيل أى حالات وفاة.

الخلاصة: فى حالات العيوب الخلقية المعقدة بالقلب التى احتاجت إلى إجراء وصلة جلين ثنائية الاتجاه ولم تتمكن هذه التحويله من تحسين نسب الأكسجين بالدم بالشكل الكافى ولديهم ما يمنع من إجراء جراحة الفونتان أو لا يمكنهم تحمل مضاعفات فونتان الواردة فإن تحويله بلالوك توسيج اليسرى تمكنت من تحسين نسب الأكسجين بالدم بشكل ملحوظ فى الحالات الواردة فى الدراسة.