

The Use Of Blalock-Taussig Shunt In Cyanotic Congenital Heart Diseases

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Abstract

Blalock-Taussig Shunt provides palliation for patients with cyanotic congenital heart diseases, where an intracardiac shunting from right-to-left with inadequate pulmonary blood flow. From June (1993) to June (2015), a retrospective study was conducted on 29 patients with complex congenital cyanotic heart diseases. They underwent Blalock-Taussig Shunt at Ibn Albitar Hospital for Cardiac Surgery. Both classical (n=4) and modified Blalock-Taussig Shunts (n=25) were done. The underlying cyanotic congenital heart diseases included tetralogy of fallot (n=14), transposition of great arteries (n=6), tricuspid atresia (n=3), double outlet ventricle (n=5) and pulmonary stenosis with atrial septal defect (n=1). The age of patients at the operation ranged between 2 weeks to 13 years with a mean age of 49 ± 59 , and the patients' weight ranged between 3-30 Kg with a mean weight of 11.3 ± 7.5 . All shunts were performed through left or right lateral thoracotomy via fourth intercostal space. In modified Blalock-Taussig Shunts, different sizes of PTFE vascular grafts ranged between 3 to 8 mm were used. Echocardiographic studies were performed for all patients, but cardiac catheterizations were less frequently used. Shunt patency and length of satisfactory palliation for all patients were determined by different criteria. The overall mortalities were 12 patients. The majority of their deaths was related to shunt failure. Early deaths were recorded for 7 patients, 5 of them were related to shunt failure, and other 2 deaths are irrelative to shunt failure. Late deaths were 5 shunts. The overall shunt failures were 12 shunts. Most of shunt failures occurred in patients with main pulmonary artery size 6mm using graft size 6mm in modified Blalock-Taussig Shunt. In conclusions, Blalock-Taussig Shunt is good palliation for cyanotic congenital heart diseases.

Key ward: Blalock-Taussig Shunt , PTFE Polytetrafluorethylene graft, palliation .

الخلاصة

توصيلة بلا لوك تستخدم كعلاج تخفيفي للمرضى الذين يعانون من تشوهات القلب الولادية الازرقاية بسبب وجود تحويلة داخل عضلة القلب لمسار الدم من الجانب الايمن الى الجانب الايسر للقلب مع نقص الدم في الشريان الرئوي. دراسة استيعادية اجريت في مستشفى ابن البيطار لجراحة القلب في بغداد تسلط الضوء على النتائج الجراحية ل 29 مريض مصابين بتشوهات القلب الولادية المعقدة والازرقاية تمت احالتهم من اقسام الباطنية لتلقي العلاج الجراحي عن طريق استخدام توصيلة بلالوك وللفترة من 01 \ 01 \ 1993 ولغاية 01 \ 12 \ 2015 حيث تم استخدام توصيلة بلالوك الكلاسيكية ل (4) مرضى فقط بينما المرضى الاخرين والبالغ عددهم (25) تم استخدام توصيلة بلالوك المعدلة حيث كان المرضى يعانون من تشوهات القلب الولادية المعقدة والازرقاية على النحو الاتي :- (14) مريض يعاني من رباعية فالوت , (6) انعكاس الشريان الرئيسية , (3) انغلاق الصمام الثلاثي , (5) بطين ثنائي المخرج , (1) تضيق الصمام الرئوي مع فتحة بين الاذنين . اعمار المرضى اثناء اجراء العملية كانت تتراوح بين اسبوعين الى ثلاثة عشر (13) سنوات بينما اوزانهم كانت تتراوح بين 3 الى 30 كيلو غرام وجميع العمليات تم اجراؤها عن طريق عملية فتح الصدر و في عملية توصيلة بلالوك المعدلة تم استخدام احجام متعددة من الوعاء المتعدد الثلاثي فلورو اثيلين الصناعي تتراوح بين (3) الى (8) ملليمتر. فحص الايكو تم اجراؤه لكل المرضى بينما فحص قسرة القلب تم استخدامه بنسب اقل حيث ان سالكية التحويلة و فترة فعالية العلاج التخفيفي لجميع المرضى تم تحديدها من خلال مواصفات مختلفة حيث كان عدد المرضى المتوفين هو (12) مريض معظمهم بسبب فشل التوصيلة لمرضى كان حجم الشريان الرئوي الرئيسي لديهم هو (6) ملليمتر مع استخدام وعاء صناعي بحجم (6) ملليمتر اثناء عملية توصيلة بلالوك المعدلة وبالرغم من ذلك فان المحصلة النهائية هي ان استخدام توصيلة بلالوك يعتبر خيار جيد في علاج تشوهات القلب الولادية المعقدة والازرقاية .

الكلمات المفتاحية :- توصيلة بلا لوك , الوعاء المتعدد الثلاثي فلورو اثيلين الصناعي , العلاج التخفيفي.

Introduction

I-1 Historical Aspects:-After 1944, when the first Blalock-Taussig operation was developed, many blue babies were saved from invalidism or death [Aubrey , 2014] . The first operation was performed on November 28, 1944, on a 15-months-old girl with the diagnosis of tetralogy of Fallot and severe pulmonary stenosis, that was published in 1945[Carl, 2013] .

I-2 Pathophysiology of CCHD:- Right-to-left shunting of systemic venous blood directly into the systemic circulation results in arterial hypoxemia and cyanosis[William G 1998].The degree of cyanosis depends on the degree of anoxia and the blood hemoglobin concentration (Hb)[Jesse E.E 1990].Cyanosis from intra cardiac shunt is unresponsive (or only minimally responsive) to an increase in the fraction of inspired oxygen. The intensity of the cyanosis is related to the volume of pulmonary blood flow. This was the rationale for palliative systemic to pulmonary artery shunts to increase pulmonary blood flow[Sabiston 2015].Clinical features of chronic cyanosis are much less commonly seen now than they were before because of early diagnosis and effective surgical treatment that became available for most forms of cyanotic congenital heart disease[William G.,1998]. A decrease in exercise tolerance, with dyspnea on exertion, is a characteristic feature of cyanotic heart disease[Keui-T 1996]. Periodic episodes of unconsciousness, termed hyper cyanotic spells, are signs of cerebral anoxia[Vobecky,1993]. Another cause of neurological injury in cyanotic children is brain abscess[Alcibar-J,1994]. Other clinical feature includes development of aortopulmonary collateral vessels failure to grow, metabolic acidosis, and respiratory distress[Sabiston, 2016].

I-3 Indications of BTS:- Any intra cardiac defect produces right-to-left shunting of unoxygenated venous blood with inadequate pulmonary circulation, to improve the pulmonary blood flow and decrease the cyanosis, if early total repair is not feasible[Mills WI 1985]. These intra cardiac defects include for example:- Tetralogy of Fallot (TOF) , Tricuspid Atresia (T.Atr.) and Pulmonary Atresia (P.Atr.) [Steven ,1996].

I-4 The Principles of Blalock – Taussig Shunt (BTS):-Blalock described the procedure as an anastomosis of the end of the divided subclavian artery to the side of the pulmonary artery. The classical shunt is generally done on the side of the chest containing the innominate artery; because the aortic arch and the innominate artery are always opposite to each other[Stewart , 1988]. Experimental studies have shown that approximately an amount of three quarters of the blood passing through a subclavian pulmonary shunt is directed to the lung on the side of the anastomosis[Pappas , 1982].

I-5 Advantages of CBTS:- It has a lower incidence of post-operative congestive heart failure and other post-operative complications evolved by central systemic – pulmonary shunt . Shunt flow is limited by the diameter of the subclavian artery [Kirklin-JW 2012]. The shunt may grow with the child, often allowing many years of palliation , Distortion of the pulmonary artery is uncommon[Rebecca ,1987]. Ligation of the shunt is often easier than taking-down of central aorto pulmonary shunts at the time of corrective procedures [Deleval MR 1981].

I-6 Disadvantages of CBTS:- It requires meticulous and time consuming dissection which may be difficult to justify in a severely ill infant[Odin-J,1995] , It can distort the peripheral pulmonary artery . It can cause shunt thrombosis or inadequate flow ,injury to the phrenic nerve, ventilator dependence, and need for diaphragmatic plication[Daniel ,1990],Horner's syndrome, hemorrhage from hilar collaterals, and lymphatic leak with chylothorax . When the subclavian artery is

divided, gangrene of the arm may occur in less than 1% of cases [Tometcki-AI,1995]. Pulmonary hypertension may develop and some infants do not have suitable anatomy because of the an unusual narrow or the short subclavian artery[Sethia ,1986].

I-7 Modified Blalock-Taussig Shunt (MBTS):- With the advent of reliable prosthetic graft material, technically easier forms of systemic-to-pulmonary artery shunting as means of palliation have become popular. Moreover because patients return within 2 years for total correction, the temporary nature of palliation justifies the use of an artificial conduit that may not have optimal longevity but that does preserve the subclavian arterial supply to the arm [Karpawich ,1985]. The Great Omond Street group popularized the MBTS consisting of side to side anastomosis between subclavian and pulmonary arteries using a polytetrafluoroethylene (PTFE) interposition graft, has become increasingly popular as an alternative to the classical shunt [Turner S.1995]. After vascular control, a small longitudinal incision is made in inferior aspect of the subclavian artery and an end-to-side anastomosis completes between the artery and the PTFE graft with continuous polypropylene suture [Edmunds ,1990].

I-8 Advantages of MBTS:- It can be performed through a right or left thoracotomy or a median stemotomy , more easily in neonates than classical shunt . Greater growth and less distortion of the pulmonary artery ,graft thrombosis is uncommon, and has been successfully treated . High early patency rate with less frequent early and late graft failure ,congestive heart failure is as uncommon and preservation of the subclavian artery[Mullen ,1996].

I-9 Disadvantages of MBTS:-Because the graft is fixed in size, distortion of the pulmonary artery by the shunt can be anticipated as the patient grows, These shunts can be slightly more difficult to take down than classic Blalock-Taussig shunt, and probably should be divided rather than ligated at the time of total correction . Leakage of the serous fluid through the interstices of the fabric of the PTFE occurs in 10-15 % of patients ,false aneurysm formation ,partial or complete obstruction of the subclavian artery distal to the graft has been reported and infection of MBTS is rare[Wells ,1991].

I-10 Successful Shunts:-A successful shunt decreases cyanosis, polycythemia and symptoms with which they are associated.The murmur of functioning shunt in the early post operative period can be well heard when the bell of a stethoscope is applied to the open endotracheal tube connection briefly during a quiescent phase of respiration[Godart ,1998].

Patient and methods

Between June 1993 and June 2015 (12 years), 29 patients received Blalock-Taussig shunt as palliation for different CCHD . Four patients had CBTS, but the majority had MBTS. There were 19 males and 10 females (M/F : 1.9:1). Their age ranged between 15 days to 13 years, with a mean age of 49 ± 59 months. The patients' weights ranged between 3-30 Kg, with a mean weight of 11.3 ± 7.5 Kg.

Table (I): Patient's age and weight relative to shunt failure.

Age	No. of patients	Weight range (Kg)	Average (Kg)	Failure		Overall failure %
<1 month	1	3	3	0	0	0
1-6 months	6	4-6	4.5	4	66	14
6 month-1year	6	5-8	6.5	3	50	10
1-3 year	5	7-15	10.6	0	0	0
3-6 year	3	13-14	13.6	2	66	7
> 6 year	8	12-30	20.5	3	37	10

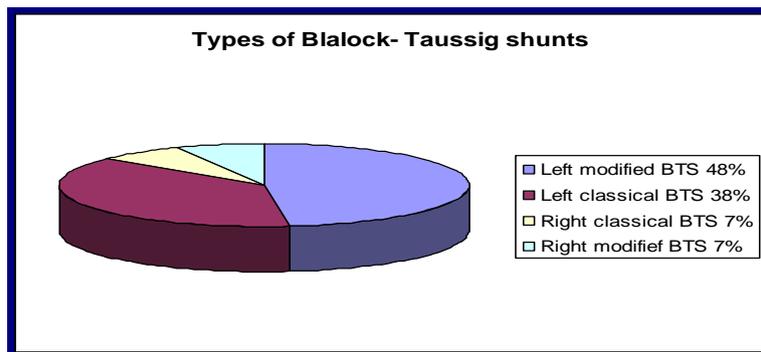


Figure (1) show types of shunts

Pre-operative diagnosis was confirmed by echocardiography and cardiac catheterization. The majority of the patients had TOF (n=14), and to less extent TGA (n=6), DOV (n=5), T.at (n=3), and severe PS with ASD (n=1). The aortic arch was left sided in 27 patients and right sided in two patients. The size of the main pulmonary artery ranges between atretic to 17mm in diameter, as reported by echocardiography.

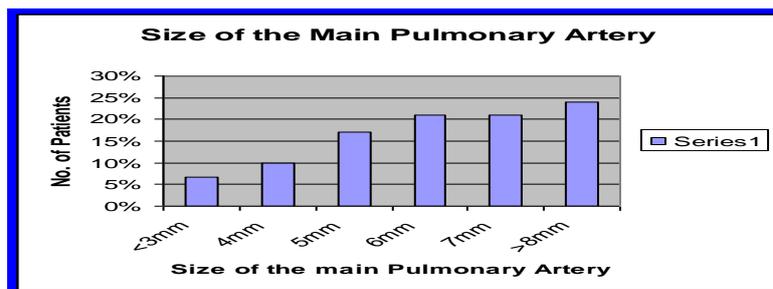


Figure (2) size of main pulmonary artery

The clinical data were collected retrospectively from patients' records. Cyanotic spells were present in all patients, with growth retardation, and recurrent chest infections in 25 patients. Three patients had associated history of subacute bacterial endocarditis(SBE) and one patient had acute renal failure(ARF) and ischemia of left lower limb .All patients had elevated Hb/Hct value ranging from 16/50 to 23/70, except the patient with ARF (14/43).Pre-operative cardiac catheterizations were performed for only 7 patients, among whom four patients with TOF, two with double outlet right ventricle(DORV), and one with TGA. One patient had unfunctioning MBTS that was performed two years ago. All shunts were constructed through left (n=16) or right (n=13) lateral thoractomy, via fourth intercostals space.In MBTS, a

conduit of Gore-Tex (PTFE) vascular graft was used, the size of which ranges between 3 to 8 mm. The size of the graft was selected according to the body surface area, and the diameter of the subclavian artery and pulmonary artery. One patient who was 15 days of age had bilateral thoracotomy at the same time of operation. First right thoracotomy was performed and discovered to have right-sided aortic with PDA, then left thoracotomy with MBTS had to be done.

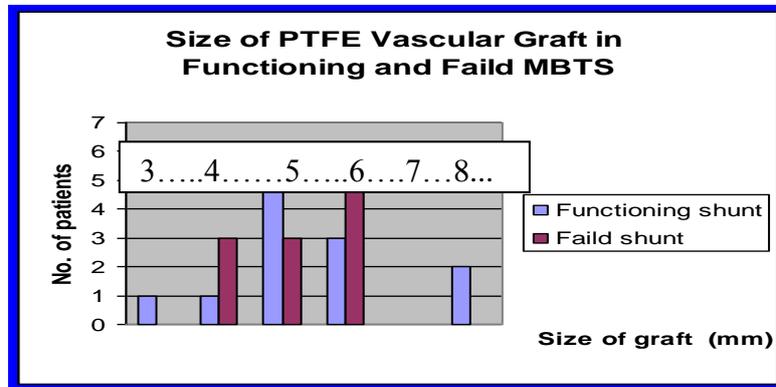


Figure (3) size of grafit in modified shunt

CBTS was performed on four patients, three of them were opposite to aortic arch and the fourth was on the same side of the aortic arch. Shunt patency was determined using a combination of clinical findings, echocardiography and cardiac catheterization. The criteria used to define that a given shunt failed to continue providing satisfactory palliation consists of any one of the followings: Progressive cyanosis with absence of continuous shunt murmur ,Occlusion of the shunt documented by echocardiography and Occlusion of the shunt documented by cardiac catheterization. The length of satisfactory palliation for any given shunt was determined to that point in time when:-(I).The shunt failed as defined above (II).The shunt was electively taken down at a complete repair. (III).Death from any reason. Patients were followed-up for a period range from 2 months to 7 years post-operatively as documented by patients record files

Results

After surgery, all surviving patients (n=28) were kept in ICU. The period of post-operative care at ICU ranged between 1-22 days. Most of the patients (17) stayed one day in ICU, including four mortalities. The other 11 patients stayed between 2-22 days. Twenty five patients received different doses of heparin ranging between 20-1000 IU/Kg/24 hours. Three patients received aspirin tablet 100mg/24 hours. The overall shunt failures were 12 shunts (41%), all of whom had MBTS. Operative mortality (within 30 days from operation) occurred in seven patients (24%), including one intra-operative death. Of the other 6 post-operative deaths, four were shunt failure related, and two had functioning shunt as documented clinically and echocardiographically.

Table (IV): Diseases and early shunt failure rates.

Diagnosis	No. of patients	Shunts		MBTS failure	
		MBTS	CBTS	No.	%
1- TOF	14	10	4	5	35
2- TGA + LVOTO	6	6	--	2	33
3- DOV + PS	5	5	--	2	40
4- T. Atr.	3	3	--	3	100
5- PS + ASD	1	1	-	--	0
Total	29	25	4	12	41

TOF Tetralogy of Fallot TGA Transposition of Great Arteries DOV Double Outlet Ventricle
 TA Tricuspid Atresia PS Pulmonary Stenosis ASD Atrial Septal Defect
 CCHD Cyanotic Congenital Heart CBTS Classical Blalock – Taussig Shunt MBTS Modified Blalock – Taussig Shunt
 DORV Double Outlet Right Ventricle PA Pulmonary Atresia PTFE Polytetrafluoroethylen
 VSD Ventricular Septal Defect Hb Hemoglobin Concentration BTS Blalock – Taussig Shunt
 DOLV Double outlet Left ventricle LVOTO Left Ventricular outflow tract Obstruction ARF acute renal failurer

Table (V): details of early shunt failure and mortality

Diagnosis	Age in months	Weight in Kg	Main pulmonary artery size mm	Graft size mm	Side of MBTS to aortic arch	Graft failure	Cause of mortality
1- TOF	4	4	5	4	Ipsilateral	+	Cardiac arrhythmia
2- TOF	48	13	6	6	Contralateral	+	Sudden cardio-pulmonary collapse
3- T.Atr. ASD, VSD	4	4	<3	4	Contralateral	+	Intra-operative death
4- T.Atr. ASD, VSD	5	6	6	5	Ipsilateral	+	Respiratory failure
5- DORV, PS	9	5	5	4	Ipsilateral	+	Respiratory failure
6- DORV, DIRV, PFO, PS, VSD	122	14.5	8	6	Ipsilateral	-	Respiratory failure
7- Severe PS, ASD, Intact IVS	2	4	4	5	Contralateral	-	Cardiac arrhythmia

Seven late shunt failures occurred during the follow-up period (24%). The graft size in those patients was 5 to 6mm. One of these patients who had associated ARF together with another three patients died at home or hospital. Two patients were still alive, and the seventh patient underwent total correction. Another late death was not related to shunt failure . Graft failure was observed more with size 6 mm.

Table (VI): Details of late shunt failures and mortalities.

Complications	No. of patients	Functioning BTS	Outcome
Bleeding	1	+	Alive
Bacterial endocarditis	3	-	2 died
Hypoxia	1	+	Alive
Chest infection	1	+	Alive
Pericardial effusion	1	-	Died
Pleural effusion	1	+	Alive
Wound infection	1	+	alive

Table (VII): Post-operative complications.

Diagnosis	Age (month)	Weight (Kg)	Main pulmonary artery size mm	Graft size mm	Post-operative failure (months)	Echo and/or cardiac catheterization	Clinical state	Results
1- TOF	56	19	6	6	25	Occluded	Progressive cyanosis	Total correction
2- TOF	90	16	6	6	40	Occluded	Progressive cyanosis	Died at hospital
3- TOF	98	16	7	6	50	Inadequate flow	- progressive cyanosis - Absence of shunt murmur	Alive
4- D. TGA	7	5	4	5	5	--	Progressive cyanosis	Died at home
5- D. TGA, VSD, PS	7	8	12	6	34	Inadequate flow	Progressive cyanosis	Died at home
6- T.Atr. ASD, PDA	144	26	7	6	62	Inadequate flow	Progressive cyanosis	Alive
7- DORV, P.S	10	8	7	5	5	Inadequate flow	- Mild cyanosis - Hypoxia	Died at hospital
8- TOF	0.5	3	3	3	35	Good flow	Mild cyanosis	Died at home

Different post-operative complications occurred, yet most of them were curable. Post-operative cardiac catheterization was performed for only 5 patients through a period ranging between 1 and 7 years. Most of these patients were without distortion of the pulmonary artery and had patent shunts.

Table (VIII): Post-operative cardiac catheterization.

Disease	Type of BTS	Post-operative catheterization date (years)	Pulmonary artery distortion	Shunt patency
D-TGA	RMBT	2	+	-
DORV	RMBT	1	-	+
TOF	LMBT	3	-	+
TOF	LMBT	2	-	+
TOF	LCBT	7	-	+

Table (II): ICU stay, shunt failure and mortality (excluding one intra-operative shunt failure and death).

Time	No. of patients	Shunt failure	mortality
1 day	17	4	4
2 days	4	--	--
3 days	3	--	1
4 days	1	--	--
> 5 days (5-22)	3	--	1
Total	28	4	6

Discussion

The use of shunting procedure in Ibn Al-Bitar Center remains limited (4.5%) as compared to others. This may be due to late referral of patients, which makes most such patients amenable for total correction of their CCHD. Many infants and children were diagnosed and sent for shunting on the basis of echocardiography only, because of its reliability, accuracy, and ease, without the need for cardiac catheterization which becomes the interventional tool which is more usable than the traditional diagnostic procedure. Similarly, in this study, cardiac catheterization was needed only for 7 patients (24%). Half of BTS were performed in patients with TOF, and it remains the commonest CCHD presented in our hospital (75%). This study included 29 patients who underwent BTS, 4 of them had CBTS and the other 25 patients had MBTS. This agrees with another study done at Saudi Arabia at Prince Sultan Cardiac Center by Dr Khalid Al Jubair and associates at 2008. It shows that 23% of patients do CBTS while 77% do MBTS in 478 patients [Khalid A. Al Jubair, 2008]. In our study, the majority of patients were over 3 months of age (n=26), and most of them had MBTS (n=23) due to late referral of patients and this disagrees with Dr. Khalid Al Jubair study which showed that the majority of patients were below 3 months of age (78 patients below one week, 270 from 1 to 12 weeks and 198 over one year of age) [Khalid A. Al Jubair, 2008].

Similarly, in this study, 13 MBTS were performed on the side of aortic arch and 12 on opposite side. Three MBTS that were constructed on ipsilateral side of aortic arch resulted in overflow to the lung and respiratory failure (23%). This may support the application of contralateral MBTS, especially for its ease to be taken down. CBTS were constructed in 4 patients, one of them was performed on the same side of aortic arch with arterioplasty of subclavian artery, because of stenotic left pulmonary artery and oligemic left lung. Following BTS, immediate increase in O₂

saturation appears post-operatively in functioning shunt. This increase was statistically significant in our study, that agrees with Al Jubair study [Khalid A. Al Jubair,2008].

In this study, different doses of heparin were given to most of patients post-operatively in ICU, and shunt failures were found to be heparin dose related. Only one patient had significant post-operative bleeding that agrees with Al Jubaier study which showed that the use of heparin decreases the shunt failure[Khalid A. Al Jubair,2008]. Out the total 29 shunts of study, 12 patients had shunt failures, according to the criteria of shunt failure. Two alive patients had progressive cyanosis, and were regarded as shunt failure, although the shunt was still patent, an inadequate flow by echo-Doppler was reported, and no further procedure was done. Thus early shunt failure rate was 41% and late shunt failure was 27% while Al jubair study recorded 9-3% of shunt failure. This high shunt failure rate in comparison to other studies may be due to the smaller size of main pulmonary artery in our study. 35% of total early shunt failures occurred in patients with TOF. All three patients with TA resulted in shunt failure. This may be due to high frequency of TOF in this study. In this study, six shunt failures occurred in MBTS with graft size 6mm and three in graft size 5mm, for equal number of patients. Yet, this was of no statistical significance. In other studies, the early operative mortality ranged between 11 % for MBTS to 8% for CBTS and most of these mortalities were not related to shunt failures[Sakai K,1987]. In this study, early mortality took place in seven patients (24%), five of them were due to shunt failure. The other two mortalities may be related to the younger age (2 months) or complex congenital cardiac pathology. Late mortality rate was 17% (n=5 patients), which is more than it was in other studies (Al jubair study show over all hospital mortality 2.9 %), and most of them were related to shunt failure (n=4). The explanation for this result may be related to poor compliance of the patient's attendance for follow up and assessment for the need for further intervention, as total correction was done for only one patient in this series. Very young age and severity of cardiac malformation have been clearly determined as a major risk factor for deaths in large series of shunting procedures. With reference to patients' age, out of the total of 12 deaths, five patients (41%) were less than 6 months of age, that agrees with Al jubair study. In this study, the palliation time within 3 years of shunting was 58%, which is comparable to other studies. The overall mean shunts survival was 26.4 ± 30.1 months, and no shunt failed between 6 and 24 months post-operatively. This indicated that shunt patency was the best up to two years (68%) from time of operation, that agrees with the study by Sakai and associated at 1987 which showed that the patency rate for 3 years range from 88.8 % for MBTS to 78 % for CBTS[33]. The mean patients' survival time with MBTS was just over 1 year for patients aged 6 months or less at operation, but it was 13 months longer for those of more than 6 months of age. These findings were nearly the same for patients 4.5 Kg or less Vs. those more than 4.5 Kg and patients with MPA size 5mm or less Vs. those more than 5mm. The size of MPA 5 mm or less appears to be the stronger risk factor than the patients' age and the size of grafts. that agrees with Al Jubair study which showed high mortality when pulmonary artery diameter was less than 4 mm. At 36 months after operation, 56% of all shunts were functioning in patients with MPA size > 5mm, 33% in MPA \leq 5mm. Another important risk factor is the very young age, as 16% of shunts were functioning in the age group 6 months or less, and 63% in the group were more than 6 months of age. In our study, 4 cases (14%) with MBTS showed distorted pulmonary artery, documented by echocardiography (n=3) and cardiac catheterization (n=1) and not related to shunt type. This agrees with Francois Godart

and associated study at 1998 . All those patients had un functioning shunts [Francois Godart,1998] . Doppler echocardiography is a highly reliable method for early post-operative evaluation of patients with BTS. Post-operative cardiac catheterization done in 17% of patients showed that the graft was functional in cases that had no pulmonary artery distortion.

Conclusion

MBTS is one of palliative procedures for CCHD .Although BTS may be indicated at any age, it is one of the higher risk in those who were less than 6 months of age . Care should be taken during shunt procedures to avoid pulmonary artery distortion, as this may lead to shunt failure ,The size of main pulmonary artery proved to be the most important determinant of BTS patency.

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