International Journal of Medical Case Reports
2022; 1(3): 29-35
http://www.sciencepublishinggroup.com/j/ijmcr
doi: 10.11648/j.ijmcr.20220103.15

Success Rate and Outcome of Stenting for the Treatment of Stenosis of Palliative Systemic-to-Pulmonary Arterial Shunts: A Case Series

Ata Firouzi[1], Ehsan Khalilipur[1], Zahra Khajali[1], Zahra Hoseini[1], Sedighe Saedi[1], Somaye Ahmadi[1, *], Atefe Shafiee[1], Amir Abdi[2]

[1]Rajaie Cardiovascular Medical and Research Center, Iran University of Medical Sciences, Tehran, Iran

[2]School of Medicine, Tehran Medical Sciences, Islamic Azad University, Tehran, Iran

Email address:

atafirouzi@yahoo.com (Ata Firouzi), Ehsankhalilipur@gmail.com (Ehsan Khalilipur), Khajaliz@yahoo.com (Zahra Khajali), Hosseini.zahra64@gmail.com (Zahra Hoseini), sedsaedi@gmail.com (Sedighe Saedi), dr_s_ahmadi@yahoo.com (Somaye Ahmadi), atefe.shafiee8968@gmail.com (Atefe Shafiee), abdiamir67@gmail.com (Amir Abdi)

[*]Corresponding author

To cite this article:

Ata Firouzi, Ehsan Khalilipur, Zahra Khajali, Zahra Hoseini, Sedighe Saedi, Somaye Ahmadi, Atefe Shafiee, Amir Abdi. Success Rate and Outcome of Stenting for the Treatment of Stenosis of Palliative Systemic-to-Pulmonary Arterial Shunts: A Case Series. *International Journal of Medical Case Reports*. Vol. 1, No. 3, 2022, pp. 29-35. doi: 10.11648/j.ijmcr.20220103.15

Received: September 2, 2022; **Accepted**: September 19, 2022; **Published**: September 29, 2022

Abstract: *Introduction:* Cyanotic congenital heart diseases includes congenital structural anomalies of the heart with shunting resulted in mixing systemic venous blood into the arterial circulation. Two types of surgical therapies are available for the treatment of this anomalies: corrective and palliative. The goal of palliative surgery is to increase the pulmonary blood flow via systemic-to-pulmonary arterial shunting as the conclusive treatment or as a bridge to corrective surgery. *Objective:* Obstruction of palliative systemic artery-to-pulmonary artery shunts is one of the causes of deterioration in the clinical status, mortality, and morbidity of patients with cyanotic heart disease. For patients contraindicated for the Fontan surgery, a solution is a new shunt insertion or interventional procedures on stenotic shunts. Redo surgery is associated with higher risks of mortality and complications. *Method:* Herein, we describe 23 patients with obstructed or stenotic systemic artery-to-pulmonary artery shunts who underwent transcatheter stenting at Rajaie Cardiovascular Medical and Research Center between 2011 and 2019. *Result:* Pulmonary blood flow increased in all the patients, and there were no significant complications or periprocedural deaths. Blalock–Taussig shunt stenting was successful in the early and late stages in all the patients with recorded data. Hematocrit was decreased by a median of 4 units (2–8 units). During the follow-up, 12 patients were rehospitalized, 4 underwent new procedures, and 4 died. *Conclusion:* Stenting an obstructed or stenotic systemic artery-to-pulmonary artery shunt is a safe and promising alternative to redo surgery with higher success rates and fewer complications compared with balloon angioplasty.

Keywords: Cyanotic Congenital Heart Disease, Palliative Systemic-to-Pulmonary Artery Shunt, Blalock–Taussig Shunt, Central Shunt, Shunt Occlusion

1. Introduction

Cyanotic congenital heart diseases, defined as any malformation of the heart presents at birth and manifests with cyanosis. Two types of surgical operations are available for the treatment of cyanotic heart diseases: corrective and palliative.

In younger infants, the rate of the primary repair of various congenital anomalies is on the rise. Nonetheless, palliative systemic artery-to-pulmonary artery shunts are still required in a significant proportion of individuals in early infancy. [1] The main goal of palliative surgery is to augment the pulmonary blood flow via systemic-to pulmonary arterial shunting or to promote the mixing of the blood through left-to-right shunting. [2] The traditional Blalock–Taussig (BT) shunt is generally acknowledged for the palliation of numerous cyanotic congenital heart diseases that need an

increase in pulmonary blood flow, either permanently or primarily as a bridge to cardiac repair or the Fontan circulation. [1, 3] Recent years have seen an increase in the use of modified BT shunts, involving the insertion of a polytetrafluoroethylene (PTFE) or GORE-TEX tube between the pulmonary artery and the subclavian artery, on the strength of their long-term success and minimal morbidity. [1] Sometimes, a modified central shunt is used to alleviate the symptoms of certain types of cyanotic heart diseases. [1] A shunt stays patent on average for 12.4 months. Generally, a 4 mm shunt becomes stenotic roughly 1 year following surgery, while a modified BT shunt meets this fate 3 years postprocedurally. [4] Because the shunt does not grow with the child, it ultimately becomes incapable of supplying enough blood to the lungs to fulfill the body's oxygen requirements. Additionally, the occlusion or stenosis of a systemic artery-to-pulmonary artery shunt, although fairly uncommon, can result in cyanosis and life-threatening hypoxemia (oxygen saturation levels <75%), necessitating prompt treatment. [1, 3, 5] If the patient's pulmonary artery size, weight, or age precludes definitive surgery, a second shunt may be necessary, which may be performed emergently to avert life-threatening complications. [1, 3].

Currently, interventional modalities such as fibrinolysis, balloon angioplasty, and coronary stent implantation for treating acute coronary syndromes are drawn upon to treat children with BT-shunt occlusion and are recommended to prevent additional surgery. [6, 7] Nevertheless, data on large populations are scarce. Indeed, whereas the majority of prior studies have described ballooning in children as case reports or short case series, [1, 2, 8, 9] only a few case reports have presented adults. [10-12].

The present study reports the outcomes of the stenting of obstructed PTFE systemic artery-to-pulmonary artery shunts in adult patients with complex cardiac disease.

2. Main Body

2.1. Method

From 2011 through 2019, a retrospective analysis was conducted on all patients (n=23) with obstructed or stenotic systemic artery-to-pulmonary artery shunts undergoing transcatheter stenting at Rajaie Cardiovascular Medical and Research Center, Tehran, Iran. The patients' demographic, clinical, echocardiographic, surgical, and interventional data were collected by reviewing computerized and paper files. Data were also obtained from existing documents on intervals between previous surgeries and interventions; critical congenital cardiac conditions; laboratory results; types and numbers of stented shunts; types, sizes, and numbers of deployed stents; and follow-ups.

The primary endpoint was procedural success, defined as the restoration of shunt patency with an increase in pulmonary blood flow through the shunt at angiography (residual stenosis of up to 20%), an elevation in the early postprocedural oxygen saturation level of 3% or more, and an improvement in functional capacity according to the New York Heart Association (NYHA) classification.

The secondary endpoints comprised periprocedural mortality, postprocedural complications, and long-term clinical outcomes such as stroke, hospital readmission, reintervention or surgery, and death.

Between 2011 and 2019, 23 patients, consisting of 16 men and 7 women, with a stenosed or occluded modified systemic artery-to-pulmonary artery shunt confirmed by cardiac catheterization and angiography were considered for shunt stenting.

Preprocedurally, the study population's arterial oxygen saturation was measured. All the patients were monitored during the procedure via pulse oximetry and electrocardiography. After local anesthesia, femoral artery and vein access was obtained. A sheath was inserted, and 50 units per kilogram weight of heparin were administered intravenously. Thereafter, hemodynamic studies were performed, followed by selective angiography of the BT shunt, the aortic root, or the descending aorta for central shunts. The narrowest diameter of the stenotic lesion in the shunt was measured in each patient. Next, subclavian arteriography or aortography was performed, and a Judkins Right catheter or a multipurpose catheter was advanced into the PTFE graft. Afterward, a soft-tip 0.035 guidewire was positioned in the pulmonary artery through the catheter, and the catheter was exchanged with a low-profile balloon catheter in 4 patients. Finally, a stent similar in size to the shunt was deployed.

A post-angioplasty angiography was performed to determine the efficacy of the procedure. A second pulse oximetry test was conducted 15 minutes after the angioplasty. The day after the procedure and 1 week thereafter, the studied patients' functional ability was measured.

Hemoglobin and hematocrit (HCT) levels were determined before and during the procedure. Additionally, the functional class of the patients was compared before and after the procedure using the NYHA system.

Between 2 months and 4 years following the procedure, the patients received primary prophylaxis with ASA and clopidogrel and were monitored for at least 1 year for death, stroke, rehospitalization, and reintervention. Follow-up data were collected from the patients' computerized and paper records, as well as interviews with them and their family members.

2.2. Results

The study population was composed of 23 patients: 16 men (69.6%) and 7 women (30.4%). The patients' ages ranged from 1 month to 27 years at the time of the initial operation, with a mean of 32±67.5 months and a median of 11.5 months (3.75–39.0 mon). Furthermore, the patients' ages varied from 15 to 40 years at the time of stenting, with a mean of 24.8±8.1 years and a median of 26.0 years (18.0–29.0 y). Additionally, the interval between surgery and attempted angioplasty was 9 months to 29 years, with a mean of 13.1±7.6 years and a median of 12.0 years (8.0–19.0 y).

Seven patients had a modified BT shunt: 1 with a classic BT shunt, 5 with bilateral modified BT shunts (all required stenting due to unilateral shunt stenosis), 1 with bilateral BT shunts and a Glenn shunt, 3 with a modified central shunt, and 1 with bilateral BT shunts and a central shunt.

The stented shunt was a right BT shunt (R-BT) in 12 patients (52.2%), of whom 1 underwent concurrent left pulmonary artery stenting. In addition, the stented shunt was a left BT shunt (L-BT) in 6 patients (26.1%), a central shunt in 3 (13%), and bilateral BT shunts in 2 (8.7%).

The Express coronary stent (Boston Scientific, Boston, MA) was implanted in 14 patients (60.8%), the Liberté coronary stent (Boston Scientific, Boston, MA) in 2 patients (8.7%), the PALMAZ BLUE coronary stent (Cordis Corporation, Bridgewater, NJ) in 2 patients (8.7%), and each of the following stents in 1 patient: the PALMAZ coronary stent (Medtronic, Minneapolis, USA), the PROMUS Element coronary stent (Boston Scientific, Natick, Massachusetts), the VALEO peripheral stent (Bard Peripheral Vascular, Tempe, Arizona, USA), the Herculink Elite renal stent (Abbott Vascular, Santa Clara, CA), the Dynamic renal stent (*BIOTRONIK*, Berlin, Germany), and the EV3 EverFlex Self-Expanding Peripheral Stent System (Medtronic, Minneapolis, USA).

Five patients (21.7%) had more than 1 stent inserted in the same shunt; all had stents of the same kind. One patient who underwent R-BT stenting with three 6 mm×18 mm Express stents and experienced immediate and sustained success with a 10-unit reduction in HCT died 7 years after the treatment because of severe heart failure. The remaining 3 patients received stenting using two 5 mm×19 mm Express stents on the L-BT, two 4.5 mm×28 mm Liberté stents on the L-BT, and two 7 mm×2 mm Blue stents on the R-BT. All 3 patients had successful procedures and are still alive.

The mean diameter of the stent was 6.0±1.0 mm, with a median of 6.0 mm (5–7 mm). Stents with a diameter of 4 mm were used in R-BTs, whereas those with a diameter of 9 mm were used in L-BTs. The median stent length was 19.0 mm, and the mean stent length was 22.6±9.2 mm.

Balloon predilation was performed on 5 patients (21.7%), all of whom experienced immediate success. In 4 patients, the success lasted for 7 days, and HCT dropped by 4 to 13 units. One patient's final success and HCT were not documented, and 2 of these patients required rehospitalization. Neither of these patients underwent another procedure or expired.

Tables 1, 2, and 3 and Picture 1 describe the patients' data.

Table 1. *Congenital heart diseases in the study population.*

Type of Congenital Heart Disease	Number
TOF	3
Pulmonary atresia +VSD	3
Tricuspid atresia	3
PS+ DTGA + VSD	2
Pulmonary atresia+ tricuspid atresia	2
Pulmonary atresia+ single ventricle	2
Pulmonary atresia+ atrophy of the pulmonary valve	1
Pulmonary atresia+ VSD+ PDA	1
DTGA+PS	1
Pulmonary atresia+ DTGA + VSD	1
DTGA	1
DTGA+ DORV	1
CCTGA+ LVSD+ DORV	1

TOF, Tetralogy of Fallot; VSD, Ventricular septal defect; DTGA, Dextro-transposition of the great arteries; PS, Pulmonary stenosis; PDA, Patent ductus arteriosus; DORV, Double-outlet right ventricle; CCTGA, Congenitally corrected transposition of the great arteries

Table 2. *Site and number of systemic artery-to-pulmonary artery shunts.*

Shunt Type	Number
Modified BT shunts	7
Bilateral modified BT shunts	5
Modified BT shunts and central shunts	5
Modified central shunts	3
Bilateral BT shunts and Glenn shunts	1
Bilateral BT shunts and central shunts	1
Classic BT shunts	1

BT, Blalock–Taussig

Table 3. *The patients data.*

	Age (y)	Primary Diagnosis	Stented Shunts	Stent Type	Predilation	Time Since Surgery (y)	Early Success	Lasting Success	Decrease in HCT	Rehospitalization	Death
1	29	VSD + pulmonary atresia	R BT	Promus 4×20	-	26	+	+	4	_	_
2	33	PS +DTGA +VSD	R BT	Benol 5×18	-	29	+	+	3	_	_
3	18	Pulmonary atresia + atrophic PV	R BT	PALMAZ Blue 7×24 (×2)	-	12	+	+	3	_	_
4	38	CCTGA+ LVSD+ DORV	R BT	Express 7×27	-	20	+	+	3	+	+
5	24	TOF + atrophic pulmonary artery	R BT	Express 7×19 & 7×15	-	1	+	+	3	_	_
6	11	DTGA+ VSD + pulmonary atresia	L BT	VALEO 9×17	+	8	+	+	5	_	_
7	29	TOF	R BT	Express 7×57	+	25	+	+	4	+	_
8	40	Tricuspid atresia	Central shunt	Express 5×15	-	9	+	+	-16	+	_
9	15	LVSD + pulmonary atresia	R+L BT	Express 5×19 (×2)	-	0.75	+	+	4	+	_

	Age (y)	Primary Diagnosis	Stented Shunts	Stent Type	Predilation	Time Since Surgery (y)	Early Success	Lasting Success	Decrease in HCT	Rehospitalization	Death
10	16	Tricuspid atresia	R BT	Liberté 5×20	-	5	+	+	8	+	_
11	27	VSD+ ASD + pulmonary atresia (TOF)	Central shunt	Express 7×27	+	12	+	+	9	_	_
12	18	DORV + DTGA	L BT	Herculink Elite 6×15	+	12	+	+	13	+	_
13	16	Tricuspid atresia + pulmonary atresia	R+L BT	Express 6×14 & 5×15	-	3	+	+	4	+	_
14	17	Pulmonary atresia + large subaortic VSD	L BT	Liberté 4.5×28 (x2)	-	8	+	+	2	+	_
15	19	TOF	LBT	PALMAZ Blue 6×24	-	15	+	+	4	+	_
16	21	Situs inversus dextrocardia + common atrium (atrioventricular concordance / ventriculoarterial discordance)	R BT	ev3 7×27	-	12	+	+	5	+	+
17	22	Situs inversus + TGA + PS + common atrium dextrocardia	L BT	Express 5×19	-	9	+	+	3	_	_
18	26	TGA + single ventricle + pulmonary atresia	R BT	Express 7×19	-	17	+	+	2	_	_
19	26	Tricuspid atresia + ASD	L BT	PALMAZ 5×18	+	19	+	+	5	_	_
20	26	DTGA + VSD + PS	Central shunt	Dynamic BT 8×38	-	9	+	+	6	+	_
21	28	Situs inversus + pulmonary atresia + VSD + PDA	R BT	Express 6×18 (X3)	-	19	+	+	10	_	_
22	33	Single ventricle + PV atresia	R BT	Express 7×19	-	13	+	+	5	_	+
23	39	Tricuspid atresia + pulmonary atresia	R BT	Express 6×18	-	18	+	+	-1	+	+

HCT, Hematocrit; TOF, Tetralogy of Fallot; BT, Blalock–Taussig; VSD, Ventricular septal defect; DTGA, Dextro-transposition of the great arteries; PS, Pulmonary stenosis; PDA, Patent ductus arteriosus; ASD, Atrial septal defect; DORV, Double-outlet right ventricle; CCTGA, Congenitally corrected transposition of the great arteries; PV, Pulmonary valve.

The treatment was well tolerated by all the patients, and stenting was completed without residual stenosis. The entire study population showed elevated pulmonary blood flow on post-angioplasty angiograms. In all the patients, BT-shunt stenting was judged effective in the early stages following the treatment. Further, long-term success was documented for all the patients by the seventh post-stenting day.

Following angioplasty, the mean oxygen saturation level in the aorta rose from 74.4%± 8.2% to 85.3%±6.7% in the studied patients. Prior to angioplasty, the median oxygen saturation level was 76% (70%–80%), and after angioplasty, it was 84.5% (82.2%–88.7%). The oxygen saturation level was enhanced by a mean of 10.4%±4.3% and a median of 10.5% (6.5%–13%).

HCT had a mean drop of 3.85 and a median decrease of 4 units (2–8 units). It was reduced by 3 or more units in 16 patients and by 2 units in 2. This number rose by 1 unit in 1 patient who had a modified R-BT shunt stented with one 6 mm×18 mm Express stent and underwent restenting 2 years later owing to restenosis. Another patient experienced an increase of 16 units in HCT and had a central shunt stented with one 5 mm×15 mm Express stent. Four years following the stenting procedure, he had central shunt implantation. According to the criteria, both of these patients had a successful angiographic operation and experienced an increase in oxygen saturation and functional ability. The others lacked HCT modifications.

Between 2 months and 4 years postprocedurally, the patients received ASA (80 mg daily) and clopidogrel (75 mg daily). There were no serious complications, including hemoptysis.

During the follow-up, 12 patients were readmitted. All of them experienced early success: 9 achieved sustained success; however, whether or not the other 3 had sustained success was unknown.

One and 4 years following the procedure, total correction was reported in 2 patients, respectively.

Five patients had redo procedures after a while:

1) A 40-year-old man with tricuspid atresia (TA) underwent central shunt stenting with one 5 mm×15

mm Express stent and experienced immediate and sustained success. Four years later, he had another central shunt implantation.

2) A 16-year-old girl with TA underwent central shunt stenting with one 5 mm×20 mm Liberté stent and experienced immediate and sustained success, with a drop in HCT of 8 units. Four years later, she had another central shunt implantation.
3) A male patient with left ventricular systolic dysfunction and pulmonary valve atresia had bilateral BT shunts. He received two 5 mm×19 mm Express stents on his L-BT and experienced early and sustained success, together with a 4-unit drop in his HCT. Seven years later, he underwent right ventricular outflow tract stenting, followed by central shunt implantation 1 year afterward.
4) An 11-year-old girl with the dextro-transposition of the great arteries, a ventricular septal defect, and pulmonary valve atresia had an L-BT shunt. She received one 9 mm ×17 mm VALEO stent, in conjunction with balloon predilation, and experienced successful early and late outcomes. She had stent ballooning a year after the stenting procedure and the Glenn shunt placement 4 years later.
5) A 39-year-old man with TA and pulmonary valve atresia had R-BT shunts. He underwent stenting with one 6 mm×18 mm Express stent and experienced immediate and sustained success, as well as a 4-unit decline in his HCT level. Seven years later, he underwent restenting.

Four deaths occurred among the study population:

1) A patient with the congenitally corrected transposition of the great arteries and the double-outlet right ventricle who underwent bilateral BT shunt implantation aged 6 months and R-BT stenting aged 38 years with one 7 mm×27 mm Express stent expired 4 years postprocedurally. The death occurred despite an effective angioplasty, increasing the arterial oxygen saturation level from 70% to 80% (sustained) and decreasing the HCT level by 3 units.
2) A patient with the dextro-transposition of the great arteries who underwent R-BT stenting with one 7 mm×27 mm EV3 EverFlex stent at the age of 21 expired 1 year postprocedurally. The death occurred despite a reported early success at 7 days' follow-up (a rise in the arterial oxygen saturation level from 77% to 85% and a 5-unit fall in the HCT level).
3) A 28-year-old man with a ventricular septal defect, a patent ductus arteriosus, and pulmonary valve atresia who underwent R-BT stenting with three 6 mm×18 mm Express stents died 1 year after the procedure. The death occurred although the patient experienced a postoperative elevation in his arterial oxygen saturation to 90% and a fall in his HCT by 10 units.
4) A 33-year-old man with a single ventricle and pulmonary valve atresia died 5 years after stenting on his R-BT with one 7 mm×19 mm Express stent. The patient died although his postoperative oxygen saturation level rose from 76% to 89%, and his functional capacity improved from NYHA class III to NYHA class I.

2.3. Discussion

Complete or even partial obstruction of palliative systemic artery-to-pulmonary artery shunts is, albeit relatively rare, a significant cause of death and morbidity in patients with shunt-dependent pulmonary blood flow due to intraluminal thrombosis, intimal hyperplasia, or suture line fibrosis. [4, 13].

In children, percutaneous angioplasty on an obstructed BT shunt offers a safe and promising alternative to redo surgery. It may enhance patients' oxygen saturation, exercise tolerance, and overall quality of life. [5] Compared with surgery, percutaneous angioplasty is associated with reduced bleeding, post-interventional wound pain, wound infection, and lengths of hospital stay. [10] Fibrinolysis, balloon angioplasty, or stent implantation can all be employed to accomplish this. [6] Follow-up angiography should be performed after balloon angioplasty to assess the outcome, rule out restenosis, and exclude vasculature disruption at the intervention site, particularly during the early postoperative period when suture lines are most susceptible. [5] Gillespie et al recommended that stent implantation be undertaken in the case of inadequate or contraindicated balloon dilatation owing to closeness to surgery. [5].

Stenting reduces the likelihood of neointimal dissection compared with balloon angioplasty on the shunt alone. [14] Kaestner et al concluded that stent implantation was a successful and long-term therapeutic option. [15].

Short- and long-term complications associated with BT-shunt stenting include in-stent thrombosis, manifested as an abrupt decline in oxygen saturation and hemodynamics, and in-stent restenosis, caused by intimal growth over time, hence the crucial significance of long-term observation. [10].

We herein described 23 patients who suffered from congenital cardiac disease and had vascular palliative central or peripheral shunts but required shunt stenting owing to shunt stenosis. Men comprised 69.6% (n=16) of the study population. The patients' ages ranged from 1 month to 27 years at the time of the initial surgery, with a mean of 32.05±67.5 months. Additionally, the patients' ages ranged from 11 to 40 years at the time of stenting, with a mean of 24.8±8.1 years, and the interval between surgery and attempted angioplasty ranged from 9 months to 29 years, with a mean of 13.1±7.6 years. The R-BT was the most often stented shunt, with 12 patients (52.2%). The Express was the most often implanted stent (in 13 patients [56.5%]), and various stents were placed in a single shunt in 5 patients (21.7%). The mean stent diameter was 6.0±1.0 mm, and the mean stent length was 22.6±9.2 mm. Five patients (21.7%) had balloon predilation. Post-angioplasty angiograms revealed that pulmonary blood flow improved in all the patients, and the treatment was well tolerated with no severe complications or periprocedural mortality. According to the criteria, BT-shunt stenting was judged effective in both the early and late stages in the entire study population. The

oxygen saturation level was enhanced by a mean of 10.4%±4.3% and a median of 10.5% (6.5%–13%). HCT was lowered by 3.85 units on average and by 4 units on a median basis (2–8 units). It was raised by 1 unit in 1 patient and by 16 units in another; both patients, by definition, had rapid and sustained success. During the follow-up, 13 patients had hospital readmission, 5 received further surgeries, and 4 died.

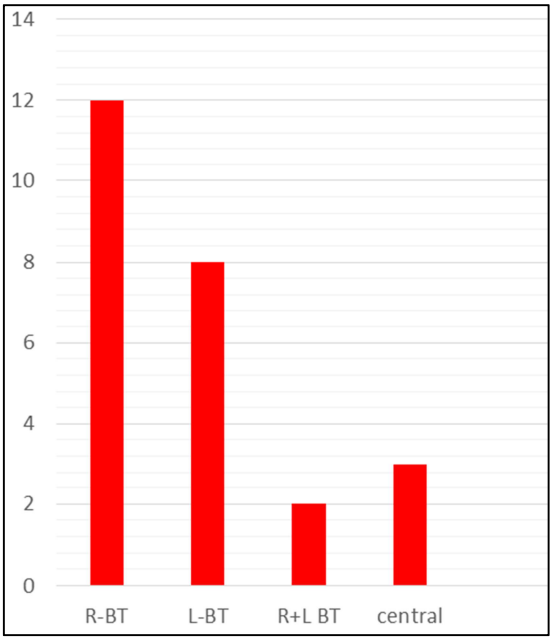
In 1989, the first successful balloon angioplasty for shunt stenosis was documented. This original description was followed by other case reports and a large retrospective series, all of which demonstrated a success rate of up to 91%. [3] In 1997, the first case report of the successful stenting of an obstructed BT shunt was published [16], followed by 7 cases of successfully implanted bare-metal, drug-eluting, or sirolimus-eluting stents in children with adequate long-term follow-ups. [10] In general, coronary artery stents (4–5 mm in diameter) are used for their trackability and modest profile (Boston Scientific, Boston, MA, Express Coronary Stent System). [5].

Between 1994 and 2014, Mathilde Bonnet et al [1] examined 28 patients (median age =0.6 y and age range =0.03–32.1 y) who underwent transcatheter intervention for the thrombotic occlusion of a modified BT shunt. Totally, 33 operations were performed, entailing 33 balloon angioplasties (100%) and 14 stent implantations (42.4%). In 6.1% of the cases, thrombolytic medications were also employed. There was no periprocedural death. Five patients (15.2%) had complications, including 1 catheter-induced transient complete atrioventricular block, 1 cardiac tamponade, and 1 major thromboembolic stroke. Early procedural success was achieved in 28 patients (84.8%) and was sustained in 26 individuals (78.8%). Procedural failure was substantially linked with young age and low body weight at the time of surgery.

From 2004 through 2008, Moszura et al [6] evaluated 23 children with occlusion or critical stenosis in the modified BT shunt. Twelve patients had local recombinant tissue plasminogen activator injections, 22 balloon angioplasties were performed, and 3 children underwent stent implantation. The procedure was effective in 22 patients (96%) and resulted in a 30% rise in arterial saturation. In 18 children, unrestricted contrast flow was achieved, whereas 3 patients had decreased central flow, and 1 kid had no flow. There were no local or systemic problems.

If shunt occlusion occurs concurrently with significant pulmonary branch stenosis, solitary stenosis angioplasty may carry the risk of additional perioperative decline in arterial saturation and mortality. Thus, in this instance, it may be more advantageous to complement percutaneous shunt recanalization with stent insertion into the stenosed pulmonary artery branches via the recanalized shunt.

Between June 1994 and May 1999, Wang et al [3] described 46 patients (age range =1 month to 7.4 years) who underwent balloon angioplasty for obstructed systemic artery-to-pulmonary artery shunts. The authors reported that 32 patients had modified BT shunts, 5 had bilateral shunts, 7 had modified central shunts, and 2 had both modified BT and central shunts. Stenosis was seen in 27 of the major pulmonary artery branches, whereas interruption occurred in 3 of these arteries. Concurrent balloon angioplasty was tried in 28 major branch pulmonary arteries, but only 25 arteries were successfully treated. Balloon dilation was efficacious in 42 patients (91%) with obstructed modified shunts, in 14 arteries with pulmonary artery stenosis, and in 11 vessels with no pulmonary artery stenosis. Oxygen saturation in the aorta rose from 74.4%±4.3% to 80.8%±3.6% following the balloon dilation angioplasty. One patient perished as a result of pneumonia. Eight patients required a modified BT shunt shortly after the surgery due to significant stenosis or interruption of the main branch pulmonary artery. After an average follow-up of 11.6 6±5.4 months, 29 patients had repeated imaging to assess the shape and size of the pulmonary arteries. Twenty-six of these 29 patients underwent open-heart surgery, with 2 mortalities.



BT: Blalock–Taussig

Figure 1. *Number of shunt types.*

3. Conclusions

In this case-series study, we described 23 patients with cyanotic congenital cardiac disease who had vascular palliative central or peripheral shunts but required shunt stenting owing to shunt stenosis. All the 23 patients well tolerated the procedure without residual stenosis and they had dramatic early and lasting improvements in the NYHA functional class and O_2 saturation, and all but 2 had decreased hemoglobin and hematocrit levels. There were no serious complications. Five patients had redo procedures after a while and 4 patients died between one and 5 years after the procedure.

Angioplasty on stenotic systemic artery-to-pulmonary artery shunts is a safer and less invasive alternative than surgery. Moreover, stenting with balloon predilation has

fewer complications and a greater success rate than balloon angioplasty alone. What kind of stents are more suitable and safer for and have better prognosis, further research is needed.

Conflict of Interests

The authors declare that they have no competing interests.

References

[1] Wang J, Wu M, Chang C, et al. Balloon angioplasty for obstructed modified systemic-pulmonary artery shunts and pulmonary artery stenoses. J Am Coll Cardiol. 2001 Mar, 37 (3) 940–947. https://doi.org/10.1016/S0735-1097(00)01194-3

[2] Zahra Hosseini, Mohammad Rafie Khorgami, Zahra Khajali. Iranian Heart Journal. 2020 Oct; 21 (4): 49-55.

[3] Bonnet M, Petit J, Lambert V, Brenot P, Riou JY, Angel CY, Belli E, Baruteau AE. Catheter-based interventions for modified Blalock-Taussig shunt obstruction: a 20-year experience. Pediatr Cardiol. 2015 Apr; 36 (4): 835-41. doi: 10.1007/s00246-014-1086-0. Epub 2015 Jan 6. PMID: 25560736.

[4] Lee ML, Chiu IS. Stent implantation for stenotic Blalock-Taussig shunts in a 5.75-year-old boy with pulmonary atresia. Int J Cardiol. 2012 Dec 15; 162 (1): e8-11. doi: 10.1016/j.ijcard.2012.04.146. Epub 2012 May 18. PMID: 22608894.

[5] Gillespie MJ, Rome JJ. Transcatheter treatment for systemic-to-pulmonary artery shunt obstruction in infants and children. Catheter Cardiovasc Interv. 2008 Jun 1; 71 (7): 928-35. doi: 10.1002/ccd.21448. PMID: 18383162.

[6] Moszura T, Zubrzycka M, Michalak KW, Rewers B, Dryzek P, Moll JJ, Sysa A, Burczynski P. Acute and late obstruction of a modified Blalock-Taussig shunt: a two-center experience in different catheter-based methods of treatment. Interact Cardiovasc Thorac Surg. 2010 May; 10 (5): 727-31. doi: 10.1510/icvts.2009.219741. Epub 2010 Feb 5. PMID: 20139195.

[7] Kogon B, Villari C, Shah N, Kirshbom P, Kanter K, Kim D, Raviele A, Vincent R. Occlusion of the modified Blalock-Taussig shunt: unique methods of treatment and review of catheter-based intervention. Congenit Heart Dis. 2007 May-Jun; 2 (3): 185-90. doi: 10.1111/j.1747-0803.2007.00095.x. PMID: 18377463.

[8] Peuster M, Fink C, Bertram H, Paul T, Hausdorf G. Transcatheter recanalization and subsequent stent implantation for the treatment of early postoperative thrombosis of modified Blalock-Taussig shunts in two children. Cathet Cardiovasc Diagn. 1998 Dec; 45 (4): 405-8. doi: 10.1002/(sici)1097-0304(199812)45:4<405::aid-ccd11>3.0.co;2-a. PMID: 9863746.

[9] Gopalakrishnan, A., Sasidharan, B., Menon, S. *et al.* Drug-eluting stent for acute Blalock-Taussig shunt thrombosis in a child—case report. *Egypt Heart J* 72, 54 (2020). https://doi.org/10.1186/s43044-020-00084-y

[10] Illner J, Reinecke H, Baumgartner H, Kaleschke G. Stenting of modified Blalock-Taussig shunt in adult with palliated pulmonary atresia and ventricular septal defect: a case report. Eur Heart J Case Rep. 2019 Nov 13; 3 (4): 1-4. doi: 10.1093/ehjcr/ytz201. PMID: 32099958; PMCID: PMC7026590.

[11] Kouatli A, Al-Ata J, Galal MO, Amin MA, Hussain A. Stent implantation to maintain patency of a stenosed Blalock Taussig shunt. Asian Cardiovasc Thorac Ann. 2005 Sep; 13 (3): 274-6. doi: 10.1177/021849230501300318. PMID: 16113004.

[12] Bader R, Somerville J, Redington A. Use of self expanding stents in stenotic aortopulmonary shunts in adults with complex cyanotic heart disease. Heart. 1999 Jul; 82 (1): 27-9. doi: 10.1136/hrt.82.1.27. PMID: 10377304; PMCID: PMC1729109.

[13] Lee KJ, Humpl T, Hashmi A, Nykanen DG, Williams WG, Benson LN. Restoration of aortopulmonary shunt patency. Am J Cardiol. 2001 Aug 1; 88 (3): 325-8. doi: 10.1016/s0002-9149(01)01654-x. PMID: 11472721.

[14] Krasemann T, Tzifa A, Rosenthal E, Qureshi SA. Stenting of modified and classical Blalock-Taussig shunts--lessons learned from seven consecutive cases. Cardiol Young. 2011 Aug; 21 (4): 430-5. doi: 10.1017/S1047951111000254. Epub 2011 Mar 17. PMID: 21411029.

[15] Kaestner M, Handke RP, Photiadis J, Sigler M, Schneider MB. Implantation of stents as an alternative to reoperation in neonates and infants with acute complications after surgical creation of a systemic-to-pulmonary arterial shunt. Cardiol Young. 2008 Apr; 18 (2): 177-84. doi: 10.1017/S1047951108001959. Epub 2008 Feb 5. PMID: 18252016.

[16] Zahn EM, Chang AC, Aldousany A, Burke RP. Emergent stent placement for acute Blalock-Taussig shunt obstruction after stage 1 Norwood surgery. Cathet Cardiovasc Diagn. 1997 Oct; 42 (2): 191-4. doi: 10.1002/(sici)1097-0304(199710)42:2<191:aid-ccd21>3.0.co;2-q. PMID: 9328706.