



Deposited via The University of Leeds.

White Rose Research Online URL for this paper:

<https://eprints.whiterose.ac.uk/id/eprint/121718/>

Version: Accepted Version

---

**Article:**

Bentham, JR, Zava, NK, Harrison, WJ et al. (2018) Duct Stenting Versus Modified Blalock-Taussig Shunt in Neonates With Duct-Dependent Pulmonary Blood Flow: Associations With Clinical Outcomes in a Multicenter National Study. *Circulation*, 137 (6). pp. 581-588. ISSN: 0009-7322

<https://doi.org/10.1161/CIRCULATIONAHA.117.028972>

---

© 2017 American Heart Association, Inc. This is an author produced version of a paper published in *Circulation*. Uploaded in accordance with the publisher's self-archiving policy.

**Reuse**

Items deposited in White Rose Research Online are protected by copyright, with all rights reserved unless indicated otherwise. They may be downloaded and/or printed for private study, or other acts as permitted by national copyright laws. The publisher or other rights holders may allow further reproduction and re-use of the full text version. This is indicated by the licence information on the White Rose Research Online record for the item.

**Takedown**

If you consider content in White Rose Research Online to be in breach of UK law, please notify us by emailing [eprints@whiterose.ac.uk](mailto:eprints@whiterose.ac.uk) including the URL of the record and the reason for the withdrawal request.

**Title: Duct stenting versus modified Blalock Taussig shunt in neonates with duct-dependent pulmonary blood flow. Associations with clinical outcomes in a multicenter national study.**

**Bentham. Duct stent or Blalock shunt for infant palliation?**

James R Bentham MD PhD<sup>1</sup>, Ngoni K Zava<sup>1</sup>, Wendy J Harrison PhD<sup>2</sup>, Arjamand Shauq MD<sup>3</sup>, Atul Kalantre MD<sup>3</sup>, Graham Derrick MD<sup>4</sup>, Robin H Chen MD<sup>4</sup>, Rami Dhillon MD<sup>5</sup>, Demetris Taliotis MD<sup>6</sup>, Sok-Leng Kang MD<sup>6</sup>, David Crossland MD<sup>7</sup>, Akintayo Adesokan MD<sup>7</sup>, Anthony Hermuzi MD<sup>7</sup>, Vikram Kudumula MD<sup>8</sup>, Sanfui Yong MD<sup>8</sup>, Patrick Noonan MD<sup>9</sup>, Nicholas Hayes MD<sup>10</sup>, Oliver Stumper MD PhD<sup>5</sup> & John DR Thomson MD<sup>1</sup>.

<sup>1</sup>Yorkshire Heart Centre, Leeds General Infirmary, Great George Street, Leeds, UK

<sup>2</sup>Leeds Institute of Cardiovascular and Metabolic Medicine, University of Leeds, UK

<sup>3</sup>Alder Hey Children's Hospital, Liverpool, UK

<sup>4</sup>Great Ormond Street Children's Hospital, London, UK

<sup>5</sup>Birmingham Children's Hospital, Birmingham, UK

<sup>6</sup>Bristol Children's Hospital, Bristol, UK

<sup>7</sup>Freeman Hospital, Newcastle, UK

<sup>8</sup>Glenfield Hospital, Leicester, UK

<sup>9</sup>Glasgow Children's Hospital, Glasgow, UK

<sup>10</sup>Wessex Heart Centre, Southampton Hospital, Southampton, UK

Correspondence to Dr JR Bentham, email: [jamie.bentham@nhs.net](mailto:jamie.bentham@nhs.net)

[Telephone number +0044 113 3927639](tel:+00441133927639)

Total word count: 3700

Keywords: Congenital heart disease, Blalock Taussig Shunt, Patent arterial duct, Ductal stent

## **Abstract**

**Background:** Infants born with cardiac abnormalities causing dependence on the arterial duct for pulmonary blood flow are often palliated with a shunt usually between the subclavian artery and either pulmonary artery. A so-called modified Blalock Taussig shunt (MBTS), allows progress through early life in order to reach an age and weight at which repair or further more stable palliation can be safely achieved. MBTSs continue to present concern for post-procedure instability and early mortality such that other alternatives continue to be explored. Duct stenting is emerging as one such alternative with potential for greater early stability and improved survival.

**Methods:** To compare post-procedure outcomes and survival to next stage palliative or reparative surgery between patients undergoing modified Blalock Taussig Shunt (MBTS) and arterial duct stenting (DS) in infants with duct-dependent pulmonary blood flow. All patients undergoing cardiac surgery and congenital interventions in the UK are prospectively recruited to an externally validated national outcome audit. From this audit, participating UK centers identified infants less than 30 days of age undergoing either a BTS or a DS for cardiac conditions with duct-dependent pulmonary blood flow between January 2012 and end December 2015. 171 patients underwent a MBTS and 83 patients an attempt at DS. Primary and secondary outcomes of survival and need for extra-corporal support (ECMO) were analyzed using multivariable logistic regression. Longer term mortality pre-repair and re-intervention were analyzed using Cox proportional hazards regression. All multivariable analyses accommodated a propensity score to balance patient characteristics between the groups.

**Results:** There was an early (to discharge) survival advantage for infants pre-next stage surgery in the DS group (OR=4.24, 95% CI 1.37 to 13.14, p=0.012). There was also a difference in the need for ECMO support post-procedure in favor of the DS group (OR=0.22, 95% CI 0.05 to 1.05, p=0.058). Longer term survival outcomes showed a reduced risk of death pre-repair in the DS group (HR=0.25, 95% CI 0.07 to 0.85, p=0.026), but a slightly increased risk of re-intervention (HR=1.50, 95% CI 0.85 to 2.64, p=0.165).

**Conclusions.** Duct stenting is a viable alternative to surgical placement of a modified Blalock-Taussig shunt with evidence for greater post-procedure stability and improved patient survival to destination surgical treatment.

## **Clinical perspective**

### **What is new?**

- Ductal stenting is a technically challenging procedure and whether it can obviate the need for a Blalock Taussig shunt (MBTS) needs to be established.
- Stenting the arterial duct is preferable over a MBTS in terms of survival to next stage surgery, early post procedure hemodynamic stability and shorter intensive care and hospital stay.
- There is a high failure rate both early, with inability to stent the duct, and late, with greater need for re-intervention on the stented duct, compared with a MBTS.

### **What are the clinical implications?**

- Stenting the arterial duct remains an important and preferred option for palliation of neonates with dependence on the arterial duct for pulmonary blood flow.
- This needs to be in the context of a well orchestrated follow up program given the high likelihood of need to re-intervene on the stented duct to provide similar length of palliation to a MBTS.

## 1 Introduction

2 Babies born with complex cardiac abnormalities are not infrequently dependent on the arterial  
3 duct for pulmonary blood flow following birth. Patency of the arterial duct can be temporarily  
4 maintained with intravenous prostaglandins whilst preparations are made for surgical palliation  
5 with a shunt. The Blalock Taussig Thomas shunt has provided palliation for congenital heart  
6 disease since the initial description in 1944<sup>1</sup>. Other than the adoption of a Gore-Tex conduit to  
7 provide a predictably-sized connection between the subclavian and pulmonary artery, the  
8 technical aspects of the procedure have remained largely unchanged over 70 years<sup>2</sup>. Whilst  
9 procedural mortality has significantly declined over time, it is still measurable and has driven  
10 consideration of alternative approaches<sup>3</sup>. This mortality largely results from early hemodynamic  
11 instability associated with diastolic run-off through the shunt and coronary artery steal. The  
12 competing alternative, first described in 1992, is a transcatheter approach to stent the arterial  
13 duct and secure pulmonary blood flow<sup>4</sup>. Given the precarious conditions we seek to palliate, it  
14 is perhaps unsurprising that stenting the arterial duct is also associated with significant  
15 morbidity and mortality<sup>5</sup>. Consequently neither procedure has emerged as superior, with  
16 preference tending to center around the chosen practice of individual programs. In the last few  
17 years there has been an increase in the number of arterial duct stent procedures performed as  
18 well as an acceptance that the difficult experiences of the early procedures have resulted in  
19 some standardization of equipment and approach<sup>6-9</sup>. It is timely and appropriate to seek to  
20 compare the two procedures given that appreciable early mortality continues to be reported  
21 for both<sup>5,10</sup>.

1 **Methods**

2 **Study population and design**

3 This was a UK multi-center cohort study of all patients less than 30 days of age with a diagnosis  
4 of duct dependent pulmonary blood flow undergoing as their first procedure either a modified  
5 Blalock Taussig shunt (MBTS) or an arterial duct stent (DS) over a four year period (1st January  
6 2012 to 31st December 2015). One-year follow up continued to 31st December 2016. Groups  
7 were assigned on an intention to treat basis; duct instrumentation regardless of whether a duct  
8 stent was successfully achieved were assigned to the DS group. All UK centers contribute to a  
9 mandatory validated prospective audit of cardiac surgical and intervention outcomes (National  
10 Congenital Heart Disease Audit) and 9 UK centers searched their audit to identify all cases that  
11 met these inclusion criteria. Written informed consent is obtained for inclusion in this audit.  
12 The primary outcome was survival to next stage surgery (either further palliation or repair),  
13 while secondary outcomes were survival to 30 days, to discharge, to one year and need for post  
14 procedure extra-corporal membrane oxygenation (ECMO). Other measures of interest included  
15 early hospital morbidity (duration of intensive care stay, need for post-procedure ventilation  
16 and hospital stay), incidence of unintended interventions pre-next stage surgery; need for  
17 pulmonary artery plasty at next stage surgery or repair and pre-repair pulmonary artery  
18 dimensions. DS procedural success was defined as stable pulmonary blood flow without need  
19 for crossover to a MBTS.

20

21 **Patient details**

1 Baseline demographics were investigated to explore similarity of the two cohorts. Pulmonary  
2 artery imaging data prior to next stage surgery were captured when performed (angiography,  
3 computed tomography or magnetic resonance imaging). Pulmonary artery dimensions were  
4 only performed if a reliable calibration factor was present and measurements were made using  
5 digital calipers. The right (RPA) and left pulmonary artery (LPA) measurements were obtained  
6 between the shunt anastomosis or duct stent insertion and proximal to the upper lobe branch  
7 pulmonary artery. Given the relative frequency of stenotic lesions, the narrowest diameter was  
8 not necessarily recorded, but rather the length of pulmonary artery most representative of the  
9 overall vessel diameter. Multiple measurements were obtained in two views and the means  
10 recorded. Pulmonary artery diameter was corrected for body surface area at the time of the  
11 procedure and also by using the Nakata index<sup>11</sup> ( $\pi\{(RPA \text{ radius}^2, \text{ mm}^2)+(LPA \text{ radius}^2,$   
12  $\text{ mm}^2)\}/4[\text{body surface area, m}^2]$ ).

13

#### 14 **Surgical and Intervention details**

##### 15 **Modified Blalock Taussig Shunt**

16 The Gore-Tex shunt (W.L. Gore and Assoc, Flagstaff, AZ) size was at the discretion of the  
17 surgeon but generally a 4mm shunt for patients over 3.5-4kg and 3.5mm shunt for weight  
18 <3.5kg. The surgical approach was at the discretion of the surgeon. The pulmonary artery end  
19 was sewn to the distal main/right or left pulmonary artery depending on anatomy with a  
20 running suture. The aortic end was sutured to either the subclavian artery or to the distal  
21 innominate artery with a similar technique.

##### 22 **Duct stent placement**

1 Access varied depending on anatomy and operator preferences. The technique has become  
2 relatively standardized to 4 French access followed by instrumentation of the duct with an  
3 0.014 inch coronary wire followed by placement of a coronary stent (mounted on a balloon  
4 between 3 and 5mm). Some operators used a 'buddy' wire to facilitate stent positioning  
5 removing this wire before deployment. In the majority of cases a 4Fr long sheath was used to  
6 provide stability and distal angiography during stent positioning. One or multiple stents were  
7 used to cover the length of the duct.

8

## 9 **Statistical analysis**

10 Stata version 14.2<sup>12</sup> was used for all statistical analyses. Baseline and univariable comparisons  
11 were performed using a Mann-Whitney comparison of non-paired samples (two-tailed) for  
12 continuous measures, and a chi-squared test or Fisher's exact test for categorical measures.  
13 Medians and interquartile ranges (IQR) are given for numeric variables, while numbers and  
14 percentages of patients are given for categorical variables. A statistical significance level of  
15  $p < 0.05$  is assumed. The presented p-values were not adjusted for multiple comparisons.

16 A directed acyclic graph (DAG)<sup>13</sup> was constructed to assess covariate causal relationships. The  
17 DAG was assessed using the R package dagitty<sup>14</sup> and found to be consistent with the dataset. A  
18 propensity score<sup>15, 16</sup> was calculated using all confounding variables: age, weight, procedure  
19 (elective or emergency), antegrade pulmonary blood flow, single ventricle status and  
20 prematurity; i.e. those variables that potentially affect both the exposure (shunt type) and the  
21 outcome (survival, ECMO or re-intervention). The balancing property was satisfied within Stata,  
22 which uses a comparison of means approach. As the propensity score cannot accommodate the

1 effects of unmeasured variables, it is assumed that there are no missing variables with sizeable  
2 causal impacts. Although patients are clustered by UK center, a multilevel analysis would not be  
3 appropriate as random effects would not be robust with only nine upper level units, hence the  
4 propensity score calculation includes the fixed effects of the centers attended. Sensitivity  
5 analysis compared the effect of including competing exposures within the calculation of the  
6 propensity score (in some instances this can improve model precision). Single-level logistic  
7 regression analysis, including propensity score, was then performed for primary and secondary  
8 outcomes, and odds ratios (OR) are reported together with 95% confidence intervals (CI). A  
9 minimally sufficient adjustment set was identified from the DAG, and a multivariable analysis  
10 that explicitly adjusted for each confounder was compared to the model which used the  
11 propensity score. Survival analysis was performed on the longer-term survival outcomes of  
12 mortality pre-repair and re-intervention, using Cox proportional hazards regression whilst  
13 accommodating the propensity score in the same manner as for the logistic regression  
14 analyses. Hazard ratios (HR) and 95% CI's are reported.

15

## 1 Results

### 2 Baseline demographics

3 Over the study period 171 neonates underwent placement of a MBTS shunt across nine centers  
4 (median age 8 days (IQR 5-15) and weight 3.1kg (IQR 2.8-3.4); table 1, figures 1 & 2,  
5 supplementary table 1). 83 neonates underwent a transcatheter procedure to place a DS  
6 (median age 8 days (IQR 4-13) and weight 3.1kg (IQR 2.8-3.5). All baseline characteristics were  
7 similar between the two groups (table 1) with similar diagnostic categorization (supplementary  
8 table 2). In the MBTS group 90% underwent a right shunt with the predominant approach being  
9 through a median sternotomy (76%). 75.6% received a 3.5mm shunt (6.1% 3mm and 18.3%  
10 4mm). DS was approached predominantly from a femoral approach (73.8%, 44 femoral artery,  
11 26 femoral vein, 12 common carotid artery [hybrid cut down and repair], 1 axillary artery). The  
12 majority of stents placed were mounted on 3.5-4mm balloons (87.1%, 24 3.5mm, 37 4mm, 6  
13 3mm and 2 4.5mm). Most cases required one or two stents (95.9%, one stent 50, two stents 20,  
14 three stents 2, and four in 1). Median procedure time was 90 minutes ( $\pm 50.4$  minutes), median  
15 fluoroscopy time 17.5 minutes ( $\pm 14.2$  minutes) and median total radiation dosage  $152\text{cGy}/\text{cm}^2$   
16 ( $\pm 340\text{ cGy}/\text{cm}^2$ ). In the DS group there were 17% failed procedures (procedural success 82.9%)  
17 requiring conversion to a MBTS (n=13). Four were for failure to cross a tortuous duct and  
18 secure adequate wire position (though procedure attempted), in two the proximal duct was not  
19 covered, in one perforation of the right ventricle occurred and in one the duct was dissected.  
20 One stent resulted in inadequate perfusion of the LPA and there were four early stent failures  
21 (occlusion, inadequate pulmonary blood flow, in-stent stenosis).

22

1 **Multivariable analysis of primary and secondary outcome measures**

2 Table 2 shows the results of the multivariable analysis, accommodating propensity score, which  
3 was calculated using all confounding variables plus the fixed effects of the center attended.  
4 Patients in the DS group had an elevated odds of surviving pre-repair, compared to patients in  
5 the MBTS group (OR=4.24, 95% CI 1.37 to 13.14, p=0.012). Patients in the DS group had  
6 reduced odds of receiving post-procedure ECMO (OR=0.22, 95% CI 0.05 to 1.05, p=0.058),  
7 compared with patients in the MBTS group. The inclusion of competing exposures in calculating  
8 propensity scores did not substantially affect the results with no improvement in the precision  
9 of estimates of effect. Analysis of the minimally sufficient adjustment set identified by the DAG  
10 gave results consistent with the propensity score analysis.

11

12 **Survival analysis of longer-term outcomes**

13 Table 3 shows the results of the survival analysis on the two longer term survival outcomes of  
14 mortality pre-repair or next stage surgery and re-intervention whilst adjusting for the  
15 propensity score. Patients in the DS group experienced a reduced risk of death pre-repair,  
16 compared to patients in the MBTS group (HR=0.25, 95% CI 0.07 to 0.85, p=0.026). This supports  
17 the result seen for survival pre-repair in the multivariable analysis. Patients in the DS group had  
18 slightly increased odds of re-intervention compared with patients in the MBTS group (HR=1.50,  
19 95% CI 0.85 to 2.64, p=0.165). The proportional hazards assumption was met for each outcome.  
20 As with the multivariable analysis, the inclusion of competing exposures in calculating the  
21 propensity score did not alter the findings. The analysis of re-intervention with the minimally  
22 sufficient adjustment set differed modestly from that using the propensity score (HR=1.96, 95%

1 CI 1.06 to 3.61,  $p=0.031$ ). In the MBTS group the majority of interstage reintervention was the  
2 need for early shunt revision or change to another source of pulmonary blood flow (11 of 15  
3 early interventions; right ventricle to pulmonary artery conduit or transannular patch) alongside  
4 later stenting of the shunt to provide a greater period of palliation (14 of 24 late interventions).  
5 In the DS group, apart from the early procedural failures that crossed over to the MBTS group  
6 interventions predominantly occurred late (23 of 30 including 14 procedures to re-stent or  
7 balloon the existing duct stent). Figures 3 and 4 show Kaplan-Meier curves comparing survival  
8 and reintervention in patients across the two groups pre-repair or next stage surgery.

9

#### 10 **Hospital and procedural morbidity**

11 The DS group had a shorter length of stay, shorter intensive care stay and fewer ventilation  
12 days (see table 4).

13

#### 14 **Pre-next stage surgery variables**

15 The DS group came to next stage palliative surgery or complete repair at a median age of 246  
16 days (IQR 176-393 days) compared with the MBTS group at a median age of 254 days (IQR 172-  
17 356 days,  $p=0.954$ ). Oxygen saturations and weight at this time point were similar across the  
18 two groups (see table 4) although hemoglobin level was higher in the MBTS group (15.6 g/dL  
19 (IQR 14.0-17.2) vs. 14.9 g/dL (IQR 12.8-16.4) in the DS group,  $p=0.027$ ). Destination surgical  
20 procedures performed were similar (supplementary table 3) with more need for pulmonary  
21 artery reconstruction work in the DS group (52% [28 of 54] vs. 39% [47 of 122],  $p=0.14$ ). Where  
22 imaging was performed (43 of 76 DS and 69 of 137 MBTS) there was no difference in the size of

- 1 the branch pulmonary arteries (DS median Nakata index  $210\text{mm}/\text{m}^2$  (IQR 166-313) vs. MBTS
- 2  $209\text{mm}/\text{m}^2$  (IQR 139-302);  $p=0.660$ ).
- 3

1 **Discussion**

2 Even in the current era Blalock Taussig shunts are associated with 30-day mortality rates far in  
3 excess of many more technically complex neonatal operations<sup>17</sup>. Post-operative instability  
4 following MBTS, with associated morbidity and mortality has resulted in a drive towards early  
5 neonatal repair where technically possible, alongside alternative approaches to secure  
6 pulmonary blood flow where the possibility of repair does not exist<sup>18, 19</sup>. In the context of  
7 avoiding high risk shunting, ductal stenting has increased in prevalence over recent years as  
8 alternative palliation in this patient group. This study analyses and compares the outcomes of  
9 ductal stenting and surgical shunting in patients with duct-dependent pulmonary blood flow in  
10 a large, unselected, prospectively collected contemporary series. This study clearly  
11 demonstrates a survival advantage with ductal stenting through to destination surgical therapy  
12 (either repair or next stage palliation). As importantly, it also demonstrates greater stability  
13 post procedure with less need for post-operative ECMO.

14  
15 Although published studies have suggested ductal stenting is an alternative to a surgical shunt,  
16 comparisons of outcomes have been limited to much smaller single center series and  
17 consequently further limited by the given institution's enthusiasm for a particular approach<sup>5, 20</sup>.  
18 Advantages and disadvantages of either approach have been difficult to demonstrate.

19  
20 These results are partly tempered by procedural success (83% for duct stenting with 17% of the  
21 group requiring an early MBTS for procedural failure). Although equipment has improved  
22 substantially since the early descriptions of the technique, complex duct anatomy remains a

23 significant technical challenge<sup>4</sup>. In addition, re-intervention in the inter-stage period is also an  
24 important issue, with 39.8% of the DS group requiring additional procedures before next stage  
25 surgery as opposed to only 24.0% of the MBTS group. Re-intervention rates in this study are not  
26 significantly different from other published single center case series<sup>20</sup> and it seems likely that  
27 achieving the length of palliation required (median of 243 days for MBTS and 231 days for DS  
28 group) comes at the cost of further transcatheter treatments in a significant proportion of  
29 patients. With this approach good palliation can be achieved with post-duct stent infants  
30 generally coming to next-stage surgery at a similar age, weight, oxygen saturations, hemoglobin  
31 and pulmonary artery dimensions to infants post-MBTS.

32

33 In the absence of randomization the major limitation and criticism for discussion is whether the  
34 two groups really are similar. The two groups reflect national UK practice and are demonstrably  
35 well-matched at baseline and continue to remain so through the study (diagnosis, single  
36 ventricle status and next stage surgery performed). However, lack of randomization is a  
37 legitimate concern. Observational analyses of this nature fail fully to account for selection bias  
38 subtly and inadvertently introduced into the study, which cannot be controlled. A simple  
39 example of selection of unknown significance would be interventionists choosing cases with  
40 echocardiographic straighter ductal courses given the higher likelihood of successful stenting.  
41 The only way to account for these factors is to perform a randomized trial of similar magnitude  
42 and design to the single ventricle reconstruction trial which compared shunt types in the  
43 Norwood procedure<sup>21</sup>. It should also be noted that assumptions and caveats are inherent  
44 within statistical analyses, and that alternative analyses could have been performed on the

45 given data. The results presented in this paper are those that the authors believe to be the  
46 most appropriate for the available data. Effect sizes may be mitigated or attenuated should  
47 other approaches be utilized.

48  
49 The main strength of this study is that it brings to the fore a national comparison of these two  
50 approaches. Given the results presented here, one reasoned approach would be to offer duct  
51 stenting to all cases in need of secure pulmonary blood flow regardless of duct morphology. We  
52 have seen no evidence that crossover from the DS group to a MBTS results in a survival  
53 disadvantage in cases where there was failure to successfully stent the duct. These infants,  
54 however, are likely to benefit from close follow-up given the high likelihood of need for re-  
55 intervention before they reach an age or weight to proceed with next stage surgery. Cost  
56 comparison of these two approaches is beyond the scope of this study but is achievable in the  
57 data presented. Less need for ECMO, shorter intensive care stay with less need for ventilation  
58 and shorter overall hospital stay need to be balanced against greater need for reintervention in  
59 the duct stent group.

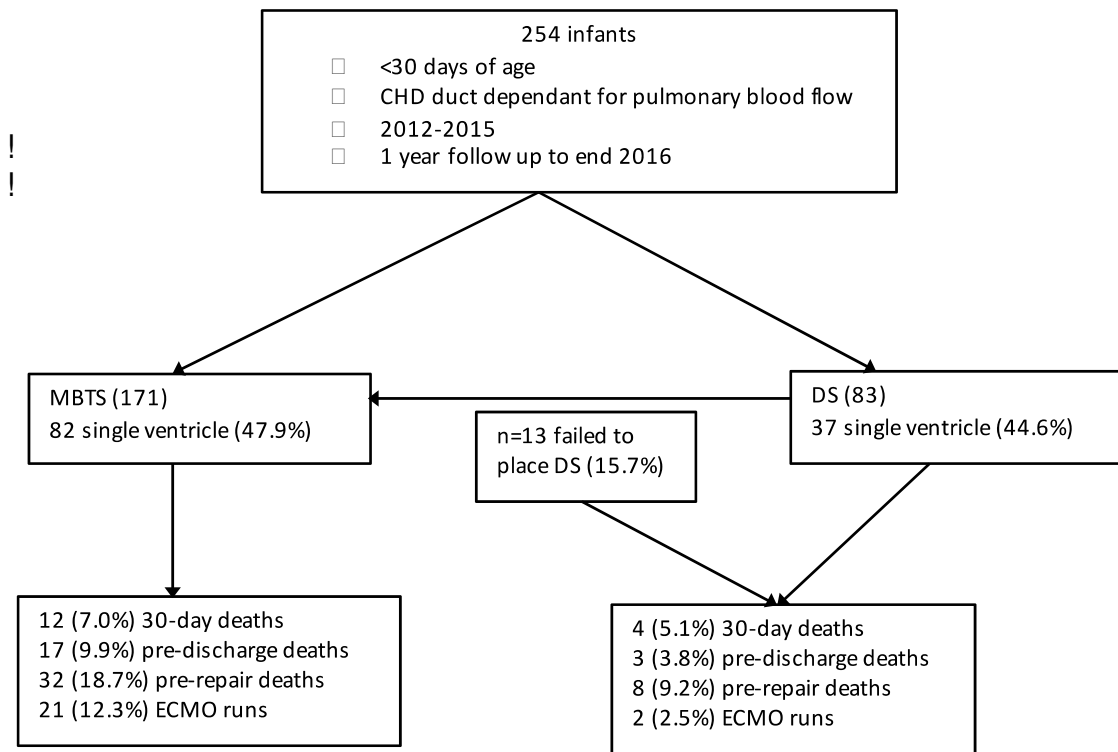
60  
61 Is stenting the arterial duct superior to a Blalock shunt? In this study, the first to assess the two  
62 competing procedures side-by-side across multiple centers, there are still twice as many shunts  
63 performed as duct stents. In the 25 years since the procedure was first described, duct stenting  
64 cannot be regarded as a panacea of palliation but we have demonstrated that it can safely be  
65 considered as a preferred alternative. This paper does offer centers confidence that this

66 procedure has tangible advantages over an arterial shunt, particularly in relation to early post-  
67 procedure stability, but the quest for truly excellent palliation remains.

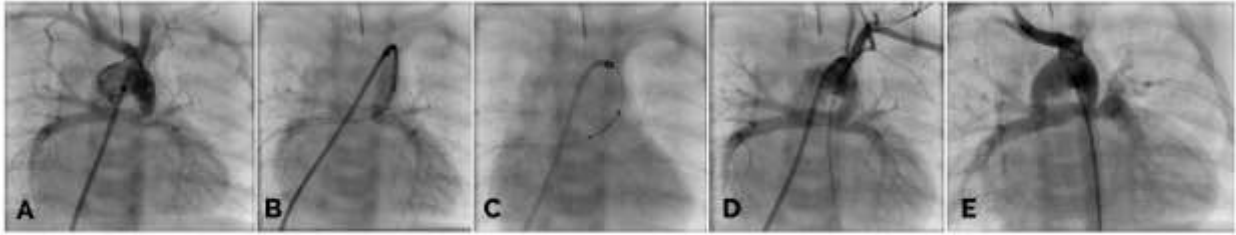
68

69 **Conclusions**

70 Stenting the arterial duct to secure pulmonary blood flow in infants with duct-dependent  
71 congenital heart disease appears to offer early survival advantage and improved early  
72 hemodynamic stability over a surgical arterial shunt. This is at the expense of procedural failure  
73 in a proportion of patients and increased likelihood of re-intervention in the inter-stage period.



**Figure 1. Study group subject selection, inclusion and exclusion criteria.** MBTS, Modified Blalock-Taussig shunt; DS, duct stent; ECMO, extra-corporal membrane oxygenation; CHD, congenital heart disease.



**Figure 2. Aortograms demonstrating duct stent placement from a femoral vein approach in a patient with pulmonary atresia ventricular septal defect. (a)** Aortogram through a four French long sheath positioned in the ascending aorta demonstrates the pulmonary artery anatomy and the insertion of the duct with restriction into the main pulmonary artery. Note the moderate left pulmonary artery narrowing which is also seen in images d and e. **(b)** two coronary wires (one working wire and one 'buddy' wire are positioned in the RPA through an internal mammary catheter in the proximal duct. **(c)** the 'buddy' wire was removed and a coronary stent has been positioned and deployed. The balloon is then removed before a repeat angiogram demonstrates good perfusion of both the right and left pulmonary vascular beds **(d)**. **(e)** Pre-repair angiogram through a pigtail catheter in the transverse arch at six months of age demonstrates acceptable growth of both pulmonary arteries. The duct stent has remained widely patent.

	MBTS	DS	Comparison
	n (%)	n (%)	P-value
Patients	171 (67.3)	83 (32.7)	-
	Median (IQR)	Median (IQR)	P-value
Age (days)	8 (5-15)	8 (4-13)	0.240
Weight (kg)	3.1 (2.8-3.4)	3.1 (2.8-3.5)	0.550
	n (%)	n (%)	P-value
Procedure			
Elective	100 (58.5)	47 (56.6)	0.100
Emergency	59 (34.5)	35 (42.2)	
Missing data	12 (7.0)	1 (1.2)	
Aortic arch			
Left	117 (68.4)	54 (65.1)	0.856
Right	27 (15.8)	15 (18.1)	
Missing data	27 (15.8)	14 (16.9)	
Antegrade PBF			
Yes	66 (38.6)	34 (41.0)	0.871
No	76 (44.4)	34 (41.0)	
Missing data	29 (17.0)	15 (18.1)	
Single ventricle future			
Yes	82 (48.0)	37 (44.6)	0.098
No – biventricular	74 (43.3)	44 (53.0)	
Missing data	15 (8.8)	2 (2.4)	
Prematurity			
Yes	10 (5.8)	5 (6.0)	0.136*
No	149 (87.1)	77 (92.8)	
Missing data	12 (7.0)	1 (1.2)	
Syndromic			
Yes	11 (6.4)	8 (9.6)	0.313
No	141 (82.5)	70 (84.3)	
Missing data	19 (11.1)	5 (6.0)	
Pre-procedure infection			
Yes	13 (7.6)	1 (1.2)	0.094
No	146 (85.4)	74 (89.2)	
Missing data	12 (7.0)	8 (9.6)	

**Table 1. Patient demographics prior to DS or MBTS demonstrating no statistically significant differences between the two groups at baseline.** Median and interquartile range (IQR) or n (%). P values obtained using chi-squared test, Fisher’s exact test (indicated by \*) or Mann-Whitney comparison of non-paired samples (two-tailed). Antegrade PBF: antegrade but inadequate pulmonary blood flow from either ventricle to the lungs.

	<b>OR (95% CI)</b>	<b>P-value</b>
Primary outcome		
Survival – pre-repair	4.24 (1.37-13.14)	0.012
Secondary outcomes		
Survival – 30 day	2.22 (0.44-11.16)	0.332
Survival – pre-discharge	6.09 (0.75-49.33)	0.091
Survival – 1 year	2.04 (0.80-5.22)	0.136
ECMO	0.22 (0.05-1.05)	0.058

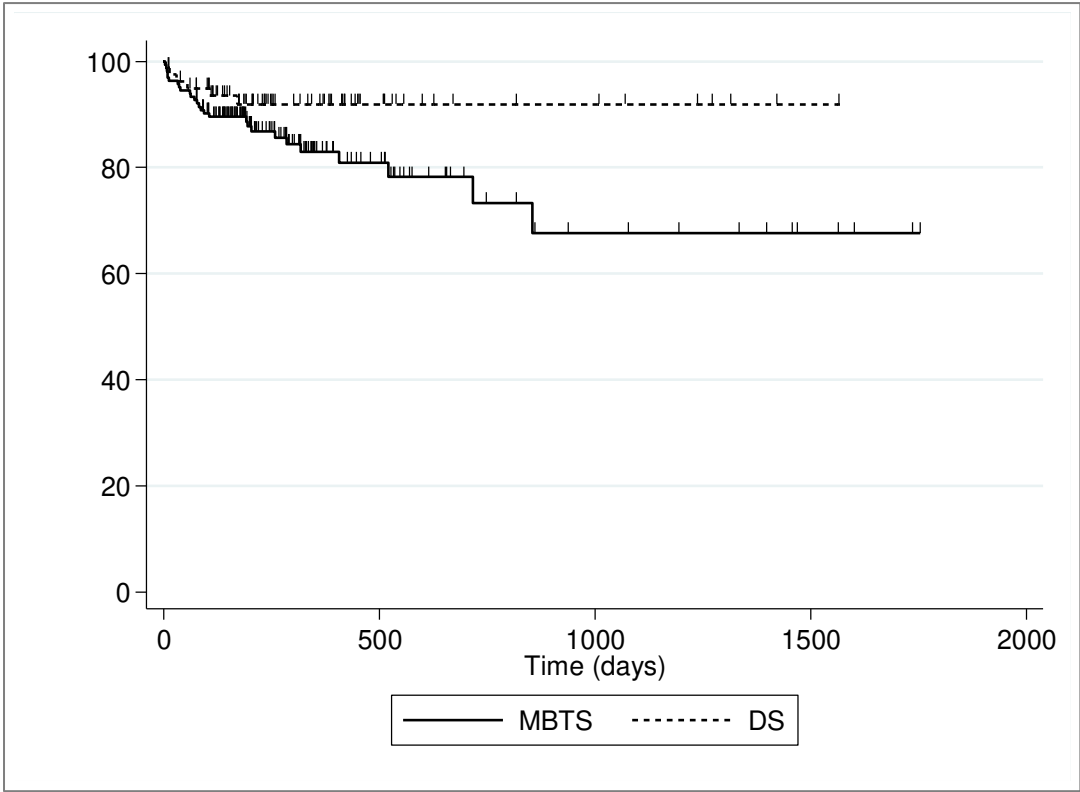
**Table 2. Multivariable analyses of primary and secondary outcomes.** OR – odds ratio of effect of DS compared with MBTS, 95% CI – confidence interval.

	<b>HR (95% CI)</b>	<b>P-value</b>
Mortality pre-repair	0.25 (0.07-0.85)	0.026
Reintervention	1.50 (0.85-2.64)	0.165

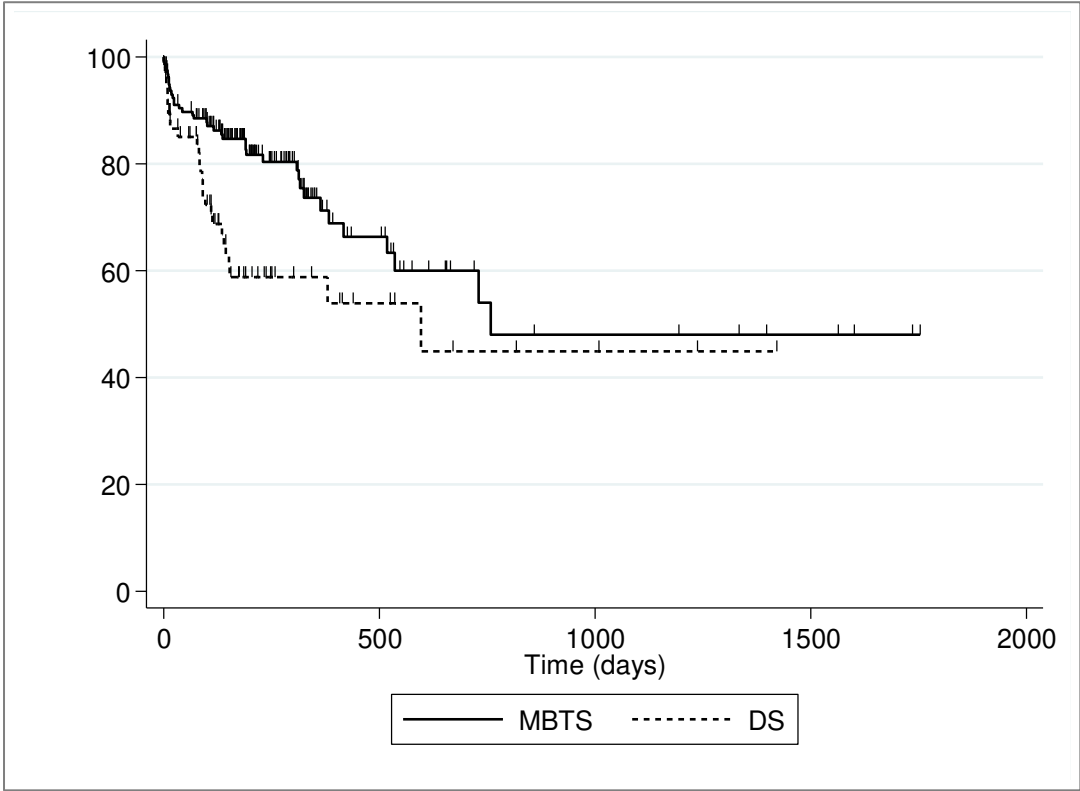
**Table 3. Survival analyses of longer term outcomes.** HR – hazard ratio of effect of DS compared with MBTS, 95% CI – confidence interval.

	<b>MBTS</b>	<b>DS</b>	<b>Comparison</b>
	<b>n (%)</b>	<b>n (%)</b>	<b>P-value</b>
Post MBTS/DS intubation			
Yes	145 (84.8)	37 (44.6)	<0.001*
No	23 (13.5)	45 (54.2)	
Missing	3 (1.8)	1 (1.2)	
Re-intervention pre-repair			
Yes	41 (24.0)	33 (39.8)	0.026*
No	126 (73.7)	49 (59.0)	
Missing	4 (2.3)	1 (1.2)	
PA plasty			
Yes	47 (27.5)	28 (33.7)	0.160
No	75 (43.9)	26 (31.3)	
Missing	49 (28.7)	29 (34.9)	
	<b>Median (IQR)</b>	<b>Median (IQR)</b>	<b>P-value</b>
LOS hospital (days)	21 (14-31)	14 (7-22)	<0.001
LOS ICU (days)	7 (4-15)	2 (0-6)	<0.001
LOS on ventilation (days)	4 (2-8)	1 (0-4)	<0.001
O <sub>2</sub> Sats post-palliation (%)	85 (80-88)	87 (82-91)	0.002
O <sub>2</sub> Sats pre-repair (%)	77 (73-81)	80 (74-84)	0.214
Weight pre-repair (kg)	7.2 (5.9-8.7)	7.0 (6.0-8.7)	0.867
Hb (g/dL)	15.6 (14.0-17.2)	14.9 (12.8-16.4)	0.027
Time to repair (days)	243 (160-351)	231 (141-380)	0.559
Age at repair (days)	254 (172-356)	246 (176-393)	0.954
LPA (mm)	6.6 (5.0-8.3)	6.1 (5.1-7.1)	0.465
RPA (mm)	6.9 (5.5-9.0)	7.0 (6.0-8.6)	0.505
Nakata index (mm <sup>2</sup> /m <sup>2</sup> )	208.9 (139.2-301.5)	210.2 (165.7-313.3)	0.660

**Table 4. Other measures of interest following DS or MBTS procedures.** Median and interquartile range (IQR) or n (%). P values obtained using chi-squared test, Fisher's exact test (indicated by \*) or Mann-Whitney comparison of non-paired samples (two-tailed). PA, pulmonary artery; LOS, length of stay; ICU, intensive care unit; LPA, left pulmonary artery; RPA, right pulmonary artery.



**Figure 3.** Kaplan-Meier curve comparing survival in patients with a modified Blalock-Taussig shunt to a duct stent to maintain adequate pulmonary blood flow to repair.



**Figure 4.** Kaplan-Meier curve comparing freedom from re-intervention in patients with a modified Blalock-Taussig shunt to a duct stent to maintain adequate pulmonary blood flow before further palliative surgery or repair.

## REFERENCES

1. Blalock A and Taussig HB. Landmark article May 19, 1945: The surgical treatment of malformations of the heart in which there is pulmonary stenosis or pulmonary atresia. By Alfred Blalock and Helen B. Taussig. *JAMA*. 1984;251:2123-38.
2. de Leval MR, McKay R, Jones M, Stark J and Macartney FJ. Modified Blalock-Taussig shunt. Use of subclavian artery orifice as flow regulator in prosthetic systemic-pulmonary artery shunts. *The Journal of thoracic and cardiovascular surgery*. 1981;81:112-9.
3. Williams JA, Bansal AK, Kim BJ, Nwakanma LU, Patel ND, Seth AK, Alejo DE, Gott VL, Vricella LA, Baumgartner WA and Cameron DE. Two thousand Blalock-Taussig shunts: a six-decade experience. *The Annals of thoracic surgery*. 2007;84:2070-5; discussion 2070-5.
4. Gibbs JL, Rothman MT, Rees MR, Parsons JM, Blackburn ME and Ruiz CE. Stenting of the arterial duct: a new approach to palliation for pulmonary atresia. *British heart journal*. 1992;67:240-5.
5. McMullan DM, Permut LC, Jones TK, Johnston TA and Rubio AE. Modified Blalock-Taussig shunt versus ductal stenting for palliation of cardiac lesions with inadequate pulmonary blood flow. *The Journal of thoracic and cardiovascular surgery*. 2014;147:397-401.
6. Gibbs JL, Uzun O, Blackburn ME, Wren C, Hamilton JR and Watterson KG. Fate of the stented arterial duct. *Circulation*. 1999;99:2621-5.
7. Schranz D, Michel-Behnke I, Heyer R, Vogel M, Bauer J, Valeske K, Akinturk H and Jux C. Stent implantation of the arterial duct in newborns with a truly duct-dependent pulmonary circulation: a single-center experience with emphasis on aspects of the interventional technique. *Journal of interventional cardiology*. 2010;23:581-8.
8. Alwi M. Stenting the ductus arteriosus: Case selection, technique and possible complications. *Annals of pediatric cardiology*. 2008;1:38-45.
9. Alwi M, Choo KK, Latiff HA, Kandavello G, Samion H and Mulyadi MD. Initial results and medium-term follow-up of stent implantation of patent ductus arteriosus in duct-dependent pulmonary circulation. *Journal of the American College of Cardiology*. 2004;44:438-45.
10. Petrucci O, O'Brien SM, Jacobs ML, Jacobs JP, Manning PB and Eghtesady P. Risk factors for mortality and morbidity after the neonatal Blalock-Taussig shunt procedure. *The Annals of thoracic surgery*. 2011;92:642-51; discussion 651-2.
11. Nakata S, Imai Y, Takanashi Y, Kurosawa H, Tezuka K, Nakazawa M, Ando M and Takao A. A new method for the quantitative standardization of cross-sectional areas of the pulmonary arteries in congenital heart diseases with decreased pulmonary blood flow. *The Journal of thoracic and cardiovascular surgery*. 1984;88:610-9.
12. Stata statistical software: release 14 [computer program]. College Station, TX: StataCorp LP; 2015.
13. Pearl J. *Causality: models, reasoning and inference*. Cambridge, UK: Cambridge University Press; 2000.
14. Textor J, van der Zander B, Gilthorpe MS, Liškiewicz M and Ellison GTH. Robust causal inference using directed acyclic graphs: the R package 'dagitty'. *International Journal of Epidemiology*. 2016;45:1887-1894.
15. Rosenbaum PR and Rubin DB. The central role of the propensity score in observational studies for causal effects. *Biometrika*. 1983;70:41-55.
16. Rosenbaum PR and Rubin DB. Reducing bias in observational studies using subclassification on the propensity score. *Journal of the American Statistical Association*. 1984;79:516-524.
17. Nicor. National congenital heart disease audit. 2017. [https://nicor4.nicor.org.uk/chd/an\\_paeds.nsf/vwcontent/home](https://nicor4.nicor.org.uk/chd/an_paeds.nsf/vwcontent/home)

18. Gladman G, McCrindle BW, Williams WG, Freedom RM and Benson LN. The modified Blalock-Taussig shunt: clinical impact and morbidity in Fallot's tetralogy in the current era. *The Journal of thoracic and cardiovascular surgery*. 1997;114:25-30.
19. Pigula FA, Khalil PN, Mayer JE, del Nido PJ and Jonas RA. Repair of tetralogy of Fallot in neonates and young infants. *Circulation*. 1999;100:II157-61.
20. Santoro G, Capozzi G, Caianiello G, Palladino MT, Marrone C, Farina G, Russo MG and Calabro R. Pulmonary artery growth after palliation of congenital heart disease with duct-dependent pulmonary circulation: arterial duct stenting versus surgical shunt. *Journal of the American College of Cardiology*. 2009;54:2180-6.
21. Ohye RG, Sleeper LA, Mahony L, Newburger JW, Pearson GD, Lu M, Goldberg CS, Tabbutt S, Frommelt PC, Ghanayem NS, Laussen PC, Rhodes JF, Lewis AB, Mital S, Ravishankar C, Williams IA, Dunbar-Masterson C, Atz AM, Colan S, Minich LL, Pizarro C, Kanter KR, Jagers J, Jacobs JP, Krawczeski CD, Pike N, McCrindle BW, Virzi L and Gaynor JW. Comparison of shunt types in the Norwood procedure for single-ventricle lesions. *N Engl J Med*. 2010;362:1980-92.

