Received: 17 September 2018 | Accepted: 16 October 2018

DOI: 10.1111/chd.12707

SPECIAL ISSUE ARTICLE

WILEY Congenital Heart Disease

Blalock-Taussig shunt versus patent ductus arteriosus stent as first palliation for ductal-dependent pulmonary circulation lesions: A review of the literature

Dana M. Boucek MD, MSCI[1] | Athar M. Qureshi MD[2] | Bryan H. Goldstein MD[3] | Christopher J. Petit MD[4] | Andrew C. Glatz MD, MSCE[1,5]

[1]The Cardiac Center at the Children's Hospital of Philadelphia, University of Pennsylvania School of Medicine, Philadelphia, Pennsylvania

[2]The Lillie Frank Abercrombie Section of Cardiology, Texas Children's Hospital, Baylor College of Medicine, Houston, Texas

[3]The Heart Institute, Cincinnati Children's Hospital Medical Center, University of Cincinnati College of Medicine, Cincinnati, Ohio

[4]Sibley Heart Center Cardiology, Children's Healthcare of Atlanta, Emory University School of Medicine, Altanta, Georgia

[5]Center for Pediatric Clinical Effectiveness, Children's Hospital of Philadelphia, Philadelphia, Pennsylvania

Correspondence
Andrew C. Glatz, MD, MSCE, Children's Hospital of Philadelphia, 6th Floor, Main Building, 34th Street and Civic Center Boulevard, Philadelphia, PA 19104.
Email: glatz@email.chop.edu

Abstract

Background: Infants with ductal-dependent pulmonary blood flow (PBF) often undergo a palliative procedure to provide a stable source of PBF prior to definitive palliation or repair. In the current era, a surgical shunt or ductal stent is used to provide PBF. We aimed to review the current literature comparing ductal stents to surgical shunts.

Methods and Results: Four small, single-center studies and two larger multicenter studies were identified comparing ductal stent to surgical shunt. Combined, these studies showed ductal stent resulted in similar or improved pulmonary artery growth, fewer complications, shorter length of stay, less diuretic use, and improved survival compared to surgical shunt. Despite inherent minor variability among the studies, ductal stent appears to be associated with more frequent reinterventions.

Conclusions: Surgical shunts remain essential to the care of these patients, but ductal stent is a reasonable alternative, and may provide some advantages in select patients with ductal-dependent PBF.

KEYWORDS

patent ductus arteriosus stent, Blalock-Taussig shunt, cyanotic congenital heart disease

1 | INTRODUCTION

For congenital heart lesions with ductal-dependent pulmonary blood flow (PBF), an initial palliative procedure to provide a more stable source of PBF is often required prior to undergoing a more definitive palliation or repair. A surgical shunt, most commonly the modified Blalock-Taussig shunt (BTS), has been a widely used initial palliative surgical procedure to provide PBF in those with ductal-dependent pulmonary flow. Despite its widespread use, the BTS has been associated with high morbidity and mortality,[1-3] prompting alternative procedures to be sought for PBF.

In 1992, stenting of the patent ductus arteriosus (PDA) was described as an alternative to BTS placement to provide PBF, and the use of this technique has expanded considerably since that time.[4-8] Patent ductus arteriosus stenting has the advantages of being less invasive, thereby avoiding a median sternotomy or lateral thoracotomy, potentially avoiding exposure to cardiopulmonary bypass, and potentially allowing for more symmetric growth of the pulmonary arteries.[9-11] The previously touted potential disadvantages of PDA stenting include procedural complications unique to this procedure (primarily vascular access related), concerns regarding stent longevity, and potential problems related to the ductal stent during subsequent surgical procedures.[12,13]

Few studies have directly compared the PDA stent to the BTS, and most of these studies have been done at single centers, have had small numbers, and/or have only looked at specific lesions.[10,14-16] Two larger multicenter studies have recently been published comparing the PDA stent to the BTS.[17,18] The key components of the six available head-to-head studies are summarized in Table 1, and these studies will be reviewed in detail in this article. Overall, the literature

TABLE 1 Characteristics of the 6 published studies comparing ductal stenting to Blalock-Taussig or other surgical shunts in patients with ductal-dependent pulmonary blood flow

Study, year of publication	Country	Enrollment period	Number of DS patients	Number of BTS patients	Patient population	Methodology	Key findings
Santoro et al. [9,10]	Italy	2003-2009	13	14	Mixed	Single-center, retrospective cohort	No difference in PA growth, more uniform PAs with DS
Amoozgar et al. [14]	Iran	2009-2011	18	20	Mixed	Single-center, retrospective cohort	No difference in PA growth, shorter LOS with DS
McMullan et al. [16]	United States	2002-2011	13	42	Mixed	Single-center, retrospective cohort	No survival difference, no difference in time to repair, no difference in reinterventions
Mallula et al. [15]	United States	2006-2013	13	16	PA/IVS	Single-center, retrospective cohort	Decreased duration of mechanical ventilation, length of stay, acute complication, and acute reinterventions with DS, increased reinterventions postdischarge with DS
Glatz et al. [17]	United States	2008-2015 (DS), 2012-2015 (BTS)	106	251	Mixed	Multicenter, retrospective cohort. Adjusted comparison using propensity sore adjustment	[a]No mortality/unplanned reintervention difference, decreased use of diuretics and fewer complication with DS, shorter ICU LOS with DS. Fewer reinterventions with BTS.
Bentham et al. [18]	UK	2012-2015	83	171	Mixed	Multicenter, retrospective cohort. Adjusted comparison using propensity score adjustment	[a]Increased odds of survival before repair with DS, no differences in reintervention

Abbreviations: DS, ductal stent; BTS, Blalock-Taussig shunt; PA, pulmonary artery; LOS, length of stay; PA/IVS, pulmonary atresia with intact ventricular septum; ICU, intensive care unit.
[a]Results from the adjusted analysis

suggests that PDA stent palliation can be a reasonable alternative to surgical BTS in select patients, and these studies are discussed in further detail below.

2 | REVIEW OF RELEVANT PUBLICATIONS

Four small studies were previously performed comparing the PDA stent to the BTS (or other surgical shunts), and will be individually reviewed in brief in the following paragraphs. Though these studies were all single center with relatively small cohort sizes, overall they showed no disadvantage of the PDA stent compared to the BTS in terms of pulmonary artery growth, survival, or reinterventions, and some studies showed some advantages of the PDA stent. In two studies, the PDA stent was associated with shorter length of stay (LOS) than BTS. One study showed a higher rate of acute complications and reinterventions in the BTS group, and the same study showed more common postdischarge reinterventions with a shorter time interval to reintervention in the PDA stent group.

A 2009 study by Santoro et al[10] looked at pulmonary artery growth following palliation with a PDA stent compared to a BTS at a single center between 2003 and 2009. The size of each group was small with 13 patients in the PDA stent group, and 14 patients in the BTS group. Both groups experienced significant growth of the pulmonary arteries as measured by the Nakata index and McGoon ratio, with no difference between the groups in terms of overall pulmonary artery growth. The PDA stent group did, however, promote more symmetric growth of the pulmonary arteries as measured by left pulmonary artery/right pulmonary artery diameter ratio.

A 2013 study by Amoozgar et al[14] compared the safety, efficacy, complications, and short-term outcomes of neonates who underwent PDA stenting to those who underwent surgical shunt (primarily BTS) from 2009 to 2011. Like the previously discussed study, there were small numbers in this study with 18 patients in the PDA stent group, and 20 patients from a different center in the surgical shunt group. A PDA stent was successful in 15 of the 18 patients. This study showed no difference in pre- or postprocedure oxygen saturations, left pulmonary artery diameter, McGoon ratio, or Nakata index, but did show a larger right pulmonary artery diameter in the PDA stent group. The PDA stent cohort was also demonstrated to have a shorter mean hospital LOS compared to the surgical shunt cohort.

A 2014 study by McMullan et al[16] evaluated the safety and the durability of a PDA stent as an alternative to a BTS from 2002 to 2011 at a single center. The numbers in the PDA stent group were similar to other studies at 13 patients, with a slightly larger BTS group (42 patients). There was no survival difference between the PDA stent and the BTS groups. There was no difference in time to second stage palliation or definitive repair, and there was no difference in postprocedural systemic oxygen saturation. There was also no difference in the number of interval reinterventions to maintain adequate PBF, and no significant difference in the time to reintervention in those that did undergo reinterventions.

A 2015 study by Mallula et al[15] compared the PDA stent to the surgical shunt in patients with pulmonary atresia with intact ventricular septum (PA/IVS) from 2006 to 2013. There were 13 patients who underwent PDA stent and 16 patients who underwent surgical shunt placement (10 central and 6 modified BTS). The baseline characteristics were similar between the 2 groups except for the PDA stent group being larger at the time of intervention. The PDA stent group had a shorter duration of mechanical ventilation, and lower median hospital LOS. Acute complications and acute reinterventions were more common in the surgical shunt group; however, postdischarge reinterventions were more common in the PDA stent group, along with a shorter time to reintervention. Although valuable, these four studies all have important limitations due to their single center nature, small cohort sizes, and nonrandomized assignment to treatment strategies. In these circumstances, there is a substantial risk of bias due to confounding by indication, whereby differences in patient characteristics influence both the assignment to treatment strategy and the outcomes of interest.

More recently, there have been 2 larger, multicenter studies published comparing the PDA stent to BTS: one U.S. study, and one British study. Because of their size and multicenter nature, these studies were able to make adjusted comparisons of outcomes to account for differences in baseline characteristics of the patients in each treatment group, in an attempt to mitigate bias due to confounding by indication. Neither study utilized a prospective randomized assignment to palliative strategy, however. Both studies showed that the PDA stent is a reasonable alternative to the BTS palliation in select patients. These two studies are reviewed in greater detail below.

The first is a U.S. study published by Glatz et al in 2018 that compared the outcomes between the BTS and the PDA stent when used as palliation for infants with ductal-dependent PBF. A relatively large multicenter cohort was utilized to allow for adjustment by potential patient-level confounding factors. This was a retrospective review of patients from four centers that comprise the Congenital Catheterization Research Collaborative (CCRC) who had either a BTS (from January 2012 through November 2015) or PDA stent (from January 2008 through November 2015). The primary outcome of this study was a composite of death or unplanned reintervention for cyanosis. The secondary outcomes included the individual components of the primary outcome, other reinterventions, intensive care unit (ICU) and hospital LOS, diuretic use at hospital discharge, procedural complications, and pulmonary artery size at last follow-up prior to definitive surgical repair.

There were 106 patients in the PDA stent group and 251 patients in the BTS group. There were differences in treatment strategies across centers, and some differences in patient-level factors. The PDA stent group included a larger number of PA/IVS (44%) patients, while the BTS group included more patients with pulmonary atresia in the setting of ventricular septal defect (39%). There were more patients with expected 2-ventricle physiology (60%) and antegrade PBF (61%) in the PDA stent group. There were no differences in gestational age, birth weight, genetic syndromes, or other comorbid conditions between the groups. There was no difference

in pre-intervention ventricular function or atrioventricular valve regurgitation by echocardiography between the groups.

The unadjusted analysis favored the PDA stent group with regard to the primary composite outcome of death or unplanned reintervention for cyanosis. It also favored the PDA stent group in terms of unplanned reintervention for cyanosis when the individual components of the composite outcome were examined separately. There was less use of diuretics at discharge and shorter total hospital and ICU LOS in the PDA stent group. The PDA stent group had larger and more symmetrical pulmonary arteries at later follow-up. The BTS group was favored in terms of other reinterventions (planned reinterventions or unplanned reinterventions to treat issues other than cyanosis), which occurred more frequently in the PDA stent cohort. Other outcomes examined in the unadjusted analysis showed a difference in the type of definitive surgical repair with more patients in the BTS group undergoing a superior cavopulmonary connection. The PDA stent group was older at the time of subsequent repair with a longer time interval between initial palliation and the definitive repair, contradicting the prevailing assumption that PDA stenting is less durable than surgical shunt. There was no difference in the need for pulmonary artery plasty at the time of definitive surgical repair, or in the rate of subsequent surgical or transcatheter pulmonary artery intervention.

Importantly, this study also performed an adjusted comparison of outcomes between the groups using propensity score adjustment techniques. The elements of the propensity score represented baseline differences between the groups and included: center, expected ultimate physiology (1 or 2 ventricle), presence of antegrade PBF, diagnostic category, preintervention inotrope use, and preintervention invasive ventilation. The adjusted analysis showed no difference between the groups with regard to the primary outcome. Similarly, there was no difference between the components of the primary outcome when examined separately. However, there was less diuretic use, fewer procedural complications, and shorter ICU LOS in the PDA stent group in the adjusted analysis. Further, the PDA stent group had larger and more symmetrical pulmonary arteries at later follow-up. The BTS group continued to have a lower risk of reinterventions in the adjusted analysis.

The second large multicenter study is a 2018 United Kingdom (U.K.) study published by Bentham et al. This study included patients identified from the National Congenital Heart Disease Audit from 9 U.K. centers, < 30 days of age with ductal-dependent PBF from January 1, 2012 to December 31, 2015. The primary outcome was survival to next stage surgery (either palliation or repair). The secondary outcomes were survival to 30 days, to discharge, and to 1 year, and need for postprocedural extracorporeal membrane oxygenation (ECMO).

There were 83 patients in the PDA stent group, and 171 patients in the BTS group. PDA stenting was successful in 70 of the 83 patients. There were no baseline differences between the groups in terms of age, weight, diagnosis, or comorbidities. The unadjusted analysis in this study showed the BTS group was intubated more commonly postprocedure, had longer LOS in the hospital and ICU, had longer mechanical ventilation, had lower oxygen saturation postprocedure, and had higher hemoglobin. There were more reinterventions before repair in the PDA stent group.

This study also performed an adjusted analysis using a propensity score. The components of the propensity score in this study were age, weight, procedure (elective or emergency), antegrade PBF, single-ventricle status, and prematurity. The adjusted analysis showed that patients in the PDA stent group had increased odds of surviving before repair compared to the BTS group. The PDA stent group had decreased odds of receiving postprocedural ECMO, though this did not meet statistical significance (p valve of 0.058). The adjusted analysis showed no significant difference in reintervention between the groups.

3 | DISCUSSION

In the current era, the BTS is still widely used in patients with ductal-dependent PBF. Given the continued high morbidity and mortality associated with the BTS over time, PDA stent placement has emerged as an alternative to surgical shunt palliation. Though the BTS will undoubtedly continue to be an important option for many patients, the literature suggests that when PDA stenting is successfully performed, it is an acceptable alternative to surgical shunt placement and may, in fact, provide several advantages. The studies reviewed herein demonstrate that PDA stent is associated with lower mortality and less morbidity than surgical shunt. Further, PDA stent palliation has been shown to result in similar or improved growth of the pulmonary arteries, decreased duration of mechanical ventilation, decreased ICU LOS, and in some cases hospital LOS, decreased diuretic burden, and lower rates of acute complications. Some studies suggested that the PDA stent may be associated with a higher number of reinterventions, but this was not uniformly identified. Moreover, in at least one large study there was no difference in *unplanned* reintervention rates with either palliative strategy. It is important to note, however, that none of these studies are truly "intent to treat," and instead focus largely on outcomes of patients who successfully receive a particular treatment strategy. In these retrospective studies, it is impossible to comprehensively assess the outcomes of all patients who may have been considered for, or had an attempt at, PDA stent placement but ultimately did not undergo this procedure because of anatomic concerns, institutional bias, or technical failure. Despite this inherent limitation to the existing body of literature notwithstanding, we conclude that, although the BTS will continue to be an essential tool in the care of patients with ductal-dependent PBF, the use of a PDA stent appears to be an acceptable alternative palliative strategy in select patients, though prospective evaluation and longer term follow-up are necessary.

CONFLICT OF INTEREST

None.

AUTHOR CONTRIBUTIONS

Dana Boucek: Data collection, data analysis/interpretation, literature review, drafting the article, and approval of the article

Athar Qureshi: Critical revision of the article and approval of the article

Bryan Goldstein: Critical revision of the article and approval of the article *Christopher Petit*: Critical revision of the article and approval of the article

Andrew Glatz: Concept/design, data analysis/interpretation, data collection, drafting of the article, critical revision of the article, supervision, and approval of the article

ORCID

Bryan H. Goldstein http://orcid.org/0000-0001-8508-9523
Andrew C. Glatz http://orcid.org/0000-0003-3791-8280

REFERENCES

1. Petrucci O, O'Brien SM, Jacobs ML, Jacobs JP, Manning PB, Eghtesady P. Risk factors for mortality and morbidity after the neonatal Blalock-Taussig shunt procedure. *Ann Thorac Surg*. 2011;92(2):642-651; discussion 651–2.
2. Dorobantu DM, Pandey R, Sharabiani MT, et al. Indications and results of systemic to pulmonary shunts: results from a national database. *Eur J Cardiothorac Surg*. 2016;49(6):1553-1563.
3. O'Connor MJ, Ravishankar C, Ballweg JA, et al. Early systemic-to-pulmonary artery shunt intervention in neonates with congenital heart disease. *J Thorac Cardiovasc Surg*. 2011;142(1):106-112.
4. Udink Ten Cate FE, Sreeram N, Hamza H, Agha H, Rosenthal E, Qureshi SA. Stenting the arterial duct in neonates and infants with congenital heart disease and duct-dependent pulmonary blood flow: a multicenter experience of an evolving therapy over 18 years. *Catheter Cardiovasc Interv*. 2013;82(3):E233-E243.
5. Alwi M, Choo KK, Latiff HA, Kandavello G, Samion H, Mulyadi MD. Initial results and medium-term follow-up of stent implantation of patent ductus arteriosus in duct-dependent pulmonary circulation. *J Am Coll Cardiol*. 2004;44(2):438-445.
6. Gewillig M, Boshoff DE, Dens J, Mertens L, Benson LN. Stenting the neonatal arterial duct in duct-dependent pulmonary circulation: new techniques, better results. *J Am Coll Cardiol*. 2004;43(1):107-112.
7. Santoro G, Gaio G, Giugno L, et al. Ten-years, single-center experience with arterial duct stenting in duct-dependent pulmonary circulation: early results, learning-curve changes, and mid-term outcome. *Catheter Cardiovasc Interv*. 2015;86(2):249-257.
8. Gibbs JL, Rothman MT, Rees MR, Parsons JM, Blackburn ME, Ruiz CE. Stenting of the arterial duct: a new approach to palliation for pulmonary atresia. *Br Heart J*. 1992;67(3):240-245.
9. Santoro G, Palladino MT, Capozzi G, Iacono C, Russo MG, Calabro R. Pulmonary artery growth following arterial duct stenting in congenital heart disease with duct-dependent pulmonary circulation. *Catheter Cardiovasc Interv*. 2009;74(7):1072-1076.
10. Santoro G, Capozzi G, Caianiello G, et al. Pulmonary artery growth after palliation of congenital heart disease with duct-dependent pulmonary circulation: arterial duct stenting versus surgical shunt. *J Am Coll Cardiol*. 2009;54(23):2180-2186.
11. Santoro G, Gaio G, Capozzi G, et al. Fate of hypoplastic pulmonary arteries after arterial duct stenting in congenital heart disease with duct-dependent pulmonary circulation. *JACC Cardiovasc Interv*. 2015;8(12):1626-1632.
12. Sivakumar K, Bhagyavathy A, Coelho R, Satish R, Krishnan P. Longevity of neonatal ductal stenting for congenital heart diseases with duct-dependent pulmonary circulation. *Congenit Heart Dis*. 2012;7(6):526-533.
13. Vida VL, Speggiorin S, Maschietto N, et al. Cardiac operations after patent ductus arteriosus stenting in duct-dependent pulmonary circulation. *Ann Thorac Surg*. 2010;90(2):605-609.
14. Amoozgar H, Cheriki S, Borzoee M, et al. Short-term result of ductus arteriosus stent implantation compared with surgically created shunts. *Pediatr Cardiol*. 2012;33(8):1288-1294.
15. Mallula K, Vaughn G, El-Said H, Lamberti JJ, Moore JW. Comparison of ductal stenting versus surgical shunts for palliation of patients with pulmonary atresia and intact ventricular septum. *Catheter Cardiovasc Interv*. 2015;85(7):1196-1202.
16. McMullan DM, Permut LC, Jones TK, Johnston TA, Rubio AE. Modified Blalock-Taussig shunt versus ductal stenting for palliation of cardiac lesions with inadequate pulmonary blood flow. *J Thorac Cardiovasc Surg*. 2014;147(1):397-401.
17. Glatz AC, Petit CJ, Goldstein BH, et al. Comparison between patent ductus arteriosus stent and modified Blalock-Taussig shunt as palliation for infants with ductal-dependent pulmonary blood flow: insights from the congenital catheterization research collaborative. *Circulation*. 2018;137(6):589-601.
18. Bentham JR, Zava NK, Harrison WJ, et al. 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*. 2018;137(6):581-588.

How to cite this article: Boucek DM, Qureshi AM, Goldstein BH, Petit CJ, Glatz AC. Blalock-Taussig shunt versus patent ductus arteriosus stent as first palliation for ductal-dependent pulmonary circulation lesions: A review of the literature. *Congenital Heart Disease*. 2019;14:105–109. https://doi.org/10.1111/chd.12707