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Impact of stent of ductus arteriosus and modified Blalock–Taussig shunt on pulmonary arteries growth and second-stage surgery in infants with ductus-dependent pulmonary circulation

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Abstract

Introduction: Ducts-dependent pulmonary circulation is spectrum of congenital heart diseases that need urgent intervention to augment pulmonary blood. Systemic to pulmonary shunt is the classical surgical management. Stenting of ductus arteriosus emerged in the last 2 decades as an alternative plausible intervention.

Objectives: To evaluate and compare the short and midterm effects of PDA stenting in compared to surgically placed shunt for augmentation of pulmonary blood flow looking to pulmonary artery (PA) branches growth, oxygen saturation and suitability for second stage repair.

Methods: We conducted this prospective study in Cardiac Surgical Intensive Care Unit. Cases were divided into “stent group” and “surgical shunt” group. Results were compared between two groups regarding oxygen saturation, mechanical ventilation duration, intensive care stay, mortality and morbidity. Growth of PA branches was assessed during follow up by echocardiograph. Nakata index score was calculated by angiogram before second stage surgery and was compared between both groups.

Results: 43 patients were included. Forty-two cases were offered stent as initial management. 6/42 cases failed stenting (14%) and 3/42 (7%) required late BT shunt after PDA stenting. 10/43 cases ended up receiving BT shunt and were counted as “surgical shunt group”. Stent group (33 cases) needed less mechanical ventilation (2.08 ± 0.65 vs. 7.8 ± 4 days with p = 0.014), and less ICU stay compared with surgical shunt group (6.2 ± 1.02 vs. 14 ± 4.5 days, P = 0.009). Both groups achieved similar growth of pulmonary artery branches (p = 0.6 for Z score of left pulmonary artery and P = 0.8 for Z score for right pulmonary artery). Although “stent group” reached second stage surgery with lower O2 saturation 67.6 ± 4.6 vs. 80 ± 4.2 in “surgical shunt” group with P value = 0.0002). Majority of patients in both groups had some PA distortion and needed surgical reconstruction in main pulmonary artery or in its main branches during second stage repair. 3 cases (7.1%) died soon post stenting versus none in surgical shunt group (p value = 0.57).

Conclusions: In neonates with ductus-dependent pulmonary circulation PDA stenting can be introduced as safe first possible option to augment pulmonary blood flow with good outcome and suitable preparation for second stage palliation.

Keywords: PDA stent, Surgical aortopulmonary shunt, Pulmonary arteries growth
1. Introduction

Aortopulmonary shunt is applied to a wide range of congenital heart diseases, collectively termed “ductus-dependent pulmonary circulation,” that will manifest early in neonatal life after closure of ductus arteriosus.

Thus, placing a shunt between both circulatory vessels is a must for survival of this group of patients. Before 1992, all these patients needed to undergo surgical shunt to maintain the perfusion through stenotic or completely atretic outflow. In the past two decades, however, patent ductus arteriosus (PDA) stenting has become a reliable palliative procedure to establish a safe and quick source of pulmonary blood flow [1].

Numerous studies have reported on the safety and advantages of PDA stenting [2,3]. Although PDA stenting may have some complications, such as migration of stent, partial PDA stent coverage, inadequate or asymmetric growth of pulmonary artery (PA) branches, when compared with those resulting from surgical shunt, such as phrenic nerve injury, vocal cord palsy, or chylothorax, the former is a less invasive procedure, particularly in relation to postoperative care [1].

However, limited knowledge exists about the viability and durability of PDA stents to facilitate adequate perfusion to pulmonary circulation and to maintain adequate growth of pulmonary branches in order to enable the patients to undergo next-stage surgery with minimal distortion of PA branches. From this perspective, ductal stenting is a practical, effective, safe, and less invasive method compared with Blalock–Taussig (BT) shunt; however, long-term stent restenosis might be a concern [4].

In this study, we aimed to assess our center’s experience with PDA stenting as an alternative choice to BT shunt to help infants overcome their defect (duct-dependent pulmonary circulation), and prepare them for next-stage surgery, and discuss short- and long-term outcomes of the procedures, in particular their impact on growth and size of pulmonary branches and the need for surgical reconstruction during second-stage surgery.

2. Materials and methods

This was a prospective study performed in the Cardiac Surgical Intensive Care Unit at Prince Sultan Cardiac Center-Qassim, Qassim, Saudi Arabia between November 2012 and January 2019. After Institutional Research Board approval, we included all patients who underwent procedures for augmentation of pulmonary blood flow. Our institutional strategy is to offer PDA stenting for all patients weighing more than 2 kg as first choice. Demographic data such as age, weight, and sex were collected. Perioperative data collected included initial cardiac diagnosis, whether single or biventricular pathway, and initial measurement of pulmonary branches with its appropriate Z scores calculated based on the Pediatric Heart Network Echo Z-score Project. Data on mechanical ventilation before and after the procedure, prostaglandin E2 infusion, PDA stent or surgical shunt (size and length of stent/shunt size, use of more than one stent to cover the PDA, need for reintervention, or failed stent implantation) and postprocedure data [oxygen saturation, duration of mechanical ventilation, length of stay (LOS) in intensive care unit (ICU)] were also collected.

Cases were followed in the pediatric cardiac clinic at 3- and 6-month intervals. Position of the stent or

3. Results

Forty-three cases of duct-dependent pulmonary circulation were included with complete data. Cases were assigned to receive surgery via either biventricular pathway (n = 27) or single ventricle pathway (n = 16). Two of the patients with single ventricle had dominant right single ventricle. Of the 43 cases, 42 were initially referred for PDA stenting; the remaining case was directed to surgical shunt because of low body weight (<2 kg). Nine cases initially assigned to receive PDA stent later had to be referred to surgical shunt. As such, we had Group 1 that included 33 cases requiring PDA stenting and Group 2 that included 10 cases requiring surgical shunt (Fig. 1). Table 1 summarizes the demographic data and variables considered for both groups.

Four (9.5%) patients from the successful stent group (Group 1) died post-PDA stenting; whereas three of the four cases died early [within 4 days after the procedure (7.1%)], one died 3 months after discharge from the hospital. The three cases that had early mortality had single ventricle physiology, with their death attributed to either pulmonary circulation stealing (n = 2) or retroperitoneal bleeding (n = 2). The fourth case had out-of-hospital cardiac arrest with an undetermined cause of late death.

Patients in the PDA stent Group 1 had less duration of mechanical ventilation (2.08 ± 0.65 days), compared with those in Group 2 (7.8 ± 4 days, p = 0.014) as well as shorter LOS in ICU (6.23 ± 1.02 vs. 14.6 ± 4.5 days, respectively, p = 0.009; Table 1). One case (3%) from the successful stent group needed reintervention during hospital stay in ICU and a second stent was reimplanted to cover the entire ductus, whereas no intervention was needed for any of the cases in Group 2. One case from each group showed signs of low cardiac output because of the placement of the large shunt and needed peritoneal dialysis during ICU admission.

In Group 2 (n = 10), five patients had tortuse PDA so stent can’t be inserted and shifted to operation room for surgical shunt. One case had stent migration to the right PA and because of significant desaturation, the patient was transferred from ICU to the operation theater to undergo urgent surgical stent retrieval and shunt placement. Three patients underwent surgical shunting before second-stage palliative surgery due to marked desaturation at the age of 2, 9, and 12 months, respectively. The decision to place a surgical shunt rather than directly performing second-stage repair in these three cases was made because of borderline size of PA branches and to allow further growth of PA branches, with pulsating PA flow achieved through the surgical shunt, before undergoing next-stage surgery (Table 2).

Femoral access was used in all cases for PDA stenting. Two cases suffered from femoral artery spasm and absent foot pulses after stenting that required prolongation of anticoagulation management until improvement in pulse and recovery of foot perfusion were achieved. Heparin infusion (21.5 ± 1.3 IU/kg/h) was initiated poststent procedure in all stented cases until oral feed was established and oral aspirin was substituted.

No differences in growth of PA branches were noted between the groups. All cases achieved acceptable growth of pulmonary branches with appropriate Z score. Twenty of 43 cases underwent second-stage surgery. The successful PDA stent group had lower oxygen saturation level following second surgery compared with Group 2 (67 ± 4.6% vs. 80 ± 4.2%, p = 0.0002). The Nakata score for the successful PDA stent group was 162 ± 26, compared with 194.75 ± 47 for the failed PDA stent group (p = 0.55). Twelve of 15 patients (80%) in the
Fig. 1. Distribution of 43 cases who had successful PDA stent or failed stent. PA = pulmonary atresia; PDA = patent ductus arteriosus.

Table 1. Comparison between the failed and successful stent groups.

<table>
<thead>
<tr>
<th>Variable</th>
<th>PDA stent (n = 33)</th>
<th>BT shunt (n = 10)</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Weight (kg)</td>
<td>3.5 ± 0.15 (35)</td>
<td>3.3 ± 0.15 (6)</td>
<td>0.59</td>
</tr>
<tr>
<td>Age (d)</td>
<td>30 ± 10 (35)</td>
<td>42 ± 17 (6)</td>
<td>0.6</td>
</tr>
<tr>
<td>RPA size (mm)</td>
<td>3.6 ± 0.26 (26)</td>
<td>3.5 ± 0.5 (2)</td>
<td>0.9</td>
</tr>
<tr>
<td>Z score of RPA</td>
<td>1.6 ± 0.23 (29)</td>
<td>1.8 ± 0.8 (2)</td>
<td>0.8</td>
</tr>
<tr>
<td>LPA size (mm)</td>
<td>3.6 ± 0.22 (26)</td>
<td>3.5 ± 0.5 (2)</td>
<td>0.2</td>
</tr>
<tr>
<td>Z score of LPA</td>
<td>1.5 ± 0.25 (29)</td>
<td>2 ± 0.83 (2)</td>
<td>0.6</td>
</tr>
<tr>
<td>Stent length (mm)</td>
<td>20 ± 1.34 (32)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Stent size</td>
<td>4.06 ± 0.05 (35)</td>
<td>3.9 ± 0.08 (6)</td>
<td>0.2</td>
</tr>
<tr>
<td>Saturation postprocedure</td>
<td>84.9 ± 1.7 (32)</td>
<td>83 ± 1.9 (6)</td>
<td>0.6</td>
</tr>
<tr>
<td>MV duration (d)</td>
<td>2.08 ± 0.65 (34)</td>
<td>7.8 ± 4 (5)</td>
<td>0.014</td>
</tr>
<tr>
<td>Heparin dose (IU/kg)</td>
<td>21.5 ± 1.3 (22)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>LOS (d)</td>
<td>6.23 ± 1.02 (31)</td>
<td>14.6 ± 4.5 (5)</td>
<td>0.009</td>
</tr>
<tr>
<td>RPA size 2 (mm)</td>
<td>5.04 ± 0.45 (16)</td>
<td>3.5 ± 0.5 (3)</td>
<td>0.9</td>
</tr>
<tr>
<td>LPA size 2 mo (mm)</td>
<td>4.8 ± 0.5 (16)</td>
<td>3.4 ± 0.5 (3)</td>
<td>0.29</td>
</tr>
<tr>
<td>RPA before second stage (mm)</td>
<td>6.04 ± 0.75 (16)</td>
<td>6.2 ± 0.8 (3)</td>
<td>0.93</td>
</tr>
<tr>
<td>Z score of RPA</td>
<td>0.85 ± 0.5 (16)</td>
<td>0.6 ± 0.6 (3)</td>
<td>0.8</td>
</tr>
<tr>
<td>LPA before second stage (mm)</td>
<td>5.9 ± 0.59 (15)</td>
<td>6.67 ± 1.8 (3)</td>
<td>0.6</td>
</tr>
<tr>
<td>Z score of LPA</td>
<td>−0.65 ± 0.4 (15)</td>
<td>0.15 ± 1.3 (3)</td>
<td>0.5</td>
</tr>
<tr>
<td>Nakata index</td>
<td>162 ± 26 (13)</td>
<td>194.7 ± 48 (3)</td>
<td>0.55</td>
</tr>
<tr>
<td>Number of cases who underwent second-stage surgery</td>
<td>51% (15)</td>
<td>50% (5)</td>
<td></td>
</tr>
<tr>
<td>Age at second procedure (mo)</td>
<td>9.8 ± 1.3 (17)</td>
<td>12.5 ± 1.8 (4)</td>
<td>0.35</td>
</tr>
<tr>
<td>Weight at second procedure (kg)</td>
<td>7.8 ± 0.5 (17)</td>
<td>9.5 ± 1.3 (3)</td>
<td>0.2</td>
</tr>
<tr>
<td>Saturation at second procedure</td>
<td>67.6 ± 4.6 (10)</td>
<td>80 ± 4.2 (3)</td>
<td>0.0002</td>
</tr>
<tr>
<td>Early 28-d mortality</td>
<td>3</td>
<td>0</td>
<td>0.57</td>
</tr>
<tr>
<td>Reconstruction</td>
<td>12/15</td>
<td>3/5</td>
<td>&gt;0.99</td>
</tr>
</tbody>
</table>

BT shunt = Blalock–Taussig shunt; LOS = length of stay; LPA = left pulmonary artery; MV = mechanical ventilation; PDA = patent ductus arteriosus; RPA = right pulmonary artery.

33 in group one and 10 in group 2 while the parenthesis is presented the number of data collected and there are some patient entered the two group.
successful PDA stent group and three of five cases (60%) in the failed PDA stent group needed reconstruction of PA branches during second-stage surgery ($p = 0.78$).

4. Discussion

Patients with duct-dependent pulmonary circulation physiology need prompt treatment in the first few days of life to ensure an adequate and stable source of pulmonary blood flow. Stenting of the PDA has been proposed since the early 1990s for maintaining duc tus patency as an alternative to surgical shunt [1]. Initially published results were encouraging with high success rate, achieving up to 93% in a single-center experience over 10 years [5]. By contrast, Alwi et al. [6] reported 91% success rate, whereas we achieved an initial success rate of 85% for stenting in our study, although the success rate for PDA stent as the only management until the second-stage surgery was almost 70%.

Difficulty to approach ductus arteriosus is usually because of torturous nature and complex morphology. The ductus arteriosus is markedly varied with regard to its origin from the aorta, size and shape, length, tortuosity, and its insertion into the PA [7]. Thus, evaluation of the anatomy by echocardiography and angiogram before attempting stenting is mandatory to enable the experts to decide on the best approach. Failure of stenting or deciding not to stent is an option chosen in the catheter laboratory. In our study, ductus could not be approached in 11.5% of cases, while Alwi et al. [6] reported unsuccessful stenting in 8.9% of cases.

The need for reintervention after PDA stent may occur early during initial admission or late after discharge from hospital. During immediate management, we had one case that had stent migration and needed emergency stent retrieval with BT shunt (3%). Schranz et al. [8] reported 3.7% stent migration in their 10-year experience, whereas in the study by Vida et al. [9], 13% of patients with PDA stent underwent emergency stent retrieval and surgical shunting due to stent migration into the LPA. In this study, three of 42 (9%) patients with PDA stenting needed stent retrieval and surgical shunting during follow-up due to significant cyanosis. Glatz et al. [1] reported surgical revision of PDA stent in 50% of their patients with PDA.

Ideally, stent length should cover the entire ductus, assuming 15–20% shortening will occur upon full expansion [7]. Of the 33 stent cases (3%) in our study, only one patient was required to go back to the catheter laboratory to implant an additional stent.

In a hospital setting mortality related to surgical procedures is not infrequent. Glatz et al. [1] reported mortality rates of around 6.6% and 10.4% in patients who received stent and BT shunt, whereas Santoro et al. [5] reported in-hospital mortality rates of 17.6% and 3.6% for patients in the stent and surgical shunt groups, respectively. In our study, the mortality rate in the stent group was 7.1%, but no deaths were reported in patients who underwent BT shunting, which comprised a small group, and thus difficult to be compared with. In general, despite wide clinical experience, neonatal mortality after surgical systemic-to-pulmonary shunting remains between 5% and 9% in the past two decades, based on data from UK centers [10].


The ICU management for both PDA stenting and surgical shunt has almost similar outcomes. Glatz et al. [1] in their recent multicenter comparison between PDA stenting and BT shunt demonstrated that both duration of mechanical ventilation and LOS in ICU and in hospital are shorter in the stent group compared with the surgical shunt group, which generally correlates with our results.

One case from each group in this study showed signs of low cardiac output and required peritoneal dialysis because of acute kidney injury. Low cardiac output may be related to the nature of congenital heart disease itself and not to the stealing phenomena, although the appropriate and adequate size of stent or even the surgical shunt for each body weight is not well known [7].

Nevertheless, short-term outcomes should not be the only focus. Long-term outcomes in infants with

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**Table 2. Cases that required emergency surgical shunt poststent placement.**

<table>
<thead>
<tr>
<th>Case</th>
<th>Type of lesion</th>
<th>Age (mo)</th>
<th>O₂ saturation (%)</th>
<th>Type of surgery</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>PA VSD</td>
<td>15</td>
<td>65</td>
<td>BT shunt</td>
</tr>
<tr>
<td>2</td>
<td>PA intact septum</td>
<td>3</td>
<td>55</td>
<td>BT shunt followed by central shunt</td>
</tr>
<tr>
<td>3</td>
<td>PA VSD</td>
<td>14</td>
<td>65</td>
<td>BT shunt followed by Rastelli</td>
</tr>
</tbody>
</table>

BT shunt = Blalock–Taussig shunt; PA = pulmonary atresia; VSD = ventricular septal defect.
ductal-dependent pulmonary blood flow are more challenging. Although most published papers, which investigated pulmonary growth as the target of palliative procedure, found that PDA stenting was equal, if not superior, to surgical shunt in achieving adequate growth of PA branches, Santoro et al. [5, 12, 13] performed a series of studies to investigate the growth and symmetry of PA following stent placement or surgical shunting, and found no difference in outcome between these two procedures.

By contrast, Glatz et al. [1] reported that the stent group achieved better growth and symmetry of pulmonary branches compared with the group that underwent surgical shunting. In our study, however, we found no difference in the growth of pulmonary branches between the two groups, with both presenting adequate growth at the time of second-stage surgery. Although the Nakata score for both groups in this study was less than 200 mm²/m², we did not prevent patients from undergoing second-stage surgery as far as the Z score of their pulmonary branches were above –2 standard deviations, considering that the lower limit of Nakata score is not yet well determined. Although the Nakata score of 250 mm²/m² is generally accepted as the cutoff for surgery, Itatani et al. [14] investigated the lower limit of PA index that can be accepted in patients undergoing Fontan operations and found that only those with pulmonary index below 110 mm²/m² will have exercise intolerance and increased pressure in their Fontan circulation. Adachi et al. [15] reported that patients with pulmonary index less than 250 mm²/m² will have acceptable function after Fontan operation. However, according to some, the lowest Nakata score to preclude a patient from surgery is not yet known and they instead suggest the Lower Lobe Index (LLI) as a more informative parameter than Nakata index in selecting patients for surgery because it is not affected by iatrogenic distortion of proximal pulmonary branches which may happen following aortopulmonary shunting and can be repaired during second-stage surgery. Furthermore, LLI is more predictive in evaluating the adequacy of the pulmonary vascular tree before surgery [16].

The timing of second stage surgery is important, as the durability of stent is limited. Patients with stents should have frequent follow-ups to assess their oxygen saturation and undergo detailed echocardiography or even elective reintervention (balloon dilation or restenting) to avoid disastrous outcome and to prolong the longevity of shunt [1]. The median age at the time of corrective surgery in our successful stent group was around 9 months, although these patients experienced lower oxygen saturation, compared with the surgical shunt group, in which patients survived longer (up to 12 months with better oxygen saturation; Table 1).

Sivakumar et al. [17] studied the durability of stents in a small group of patients and concluded that a stent may be enough for single ventricle patients; however, corrective surgery should be performed earlier in patients with biventricular repair following stent placement (within 5–14 months vs. 8–15 months in single ventricle patients). Vida et al. [9] studied the surgical outcome post-PDA stent placement and found that 53% of the stented patients needed reintervention to bridge them to second-stage palliation at a mean age of 11 months with mean oxygen saturation of 79%.

Because we did not perform elective reintervention in our center, our patients had less oxygen saturation following corrective surgery in comparison with patients who underwent surgical shunting.

The exact half-life of stent remains unknown, but it has been reported that endothelialization in the mesh of stent occurs as early as 1 month following stent insertion [18]. Senator et al. [12] reported that 45% of stented PDA will close in 4 months.

Future changes in stent design are expected to reduce endothelial reaction. Some authors have reported novel drug-eluting stents coated with poly-peptide to inhibit neointimal hyperplasia and reduce vascular smooth muscle proliferation. However, the safety and efficacy of such novel stents are not proven yet [19]. For now, repeated dilation may offer a temporary solution for endothelial proliferation in the stented duct.

Because both surgical aortopulmonary shunt and PDA stent will cause some distortion to PA branches, we compared the need for additional interventions on the pulmonary arteries during second-stage surgery between the two groups. About 80% of successful stent cases in our study needed reconstruction of either main PA or its branches, whereas only 60% required reconstruction in the failed stent group. Some authors [9] reported the need for reintervention in 46% of their cases before reaching the final surgery stage, with 53% of the cases requiring PA enlargement and plasty during second-stage surgery. By contrast, in the study by Vida et al. [9], among 13 patients who received a PDA stent, nearly half required additional pulmonary arterioplasty at the time of complete surgical repair and four needed additional PA interventions at a later period.

Although PDA stents have a tendency to develop neointimal proliferation over time, which may affect their patency and durability [1], and thus patients may require reintervention in the form pulmonary
plasty, in a good number of cases, the stenting appears to be an alternative noninvasive procedure comparable to surgical systemic-to-pulmonary shunting to prepare patients for second-stage surgery.

The major limitation of this study is the lack of proper randomization with a small number of surgical shunt patients included because of our preferred approach to stent ductal-dependent pulmonary circulation rather than dividing patients randomly to receive interventional or surgical options, which biases the procedure selection.

5. Conclusion

It is reasonable to assume that PDA stenting is a growing noninvasive approach for initial palliation of ductal-dependent pulmonary circulation with acceptable outcome and smoother postprocedure care. On the mid-term follow-up, the procedure was successful in achieving its target in 70% of cases with results comparable to surgical shunt.

Conflict of interest

The authors have no conflict of interest to report.

References


