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Case Report

Stenting of Ductus Arteriosus for Duct-Dependent Pulmonary Circulation: An Emergency and Life Saving Procedure

Desy Ayu Permitasari[1], Aulia Rizki Andini[1], Anggita Rahma Ayukusuma[1], Martvera Susilawati[2], Agus Priyatno[2]

[1]Departement of Cardiology and Vascular Medicine Dokter Kariadi Hospital/ Faculty of Medicine Diponegoro University Semarang, Indonesia
[2]Departement of Pediatric Dokter Kariadi Hospital/Faculty of Medicine Diponegoro University Semarang, Indonesia

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Author Affiliation:
Departement of Cardiology and Vascular Medicine
Dokter Kariadi Hospital/Faculty of Medicine
Diponegoro University
Semarang, Indonesia

Author Correspondence:
Desy Ayu Permitasari
Dr. Sutomo Street No. 16 Semarang,
Central Java 50244, Indonesia

E-mail Address:
desyayupermitasari@gmail.com

Abstract

Background : Critical congenital heart defect with ductal-dependent pulmonary flow presents as a life-threatening condition. The patency of ductus arteriosus is required to supply blood flow into the lung. Stent implantation into ductus arteriosus has been proposed as non-surgical management for critical congenital heart disease with duct-dependent.
Case Description: We present full-term newborns who had critical congenital heart disease with ductal-dependent pulmonary blood blow and successfully managed with ductal stenting. Patient A, a 13-days-old male newborn had Tetralogy of Fallot-Pulmonary Atresia, restrictive Ventricular Septal Defect (VSD) and patent ductus arteriosus (PDA). Patient B, a 5-days-old female newborn had Tricuspid Atresia, restrictive VSD, PDA, and multiple congenital anomaly. Patient C, a 2-days-old female newborn had PA-IVS, PDA with stretched Patent Foramen Ovale (PFO). Prostaglandin-E1 infusion was started at first and stopped 6 hours prior to the procedure. All patients underwent ductus arteriosus stenting via femoral artery approach with drug-eluting stent. Pulmonary vascular markings were increased and oxygen saturations were improved in all of the patient. 5-month follow up, patients were in stable condition and prepared for surgical correction.
Discussion : Ductal stenting in patient with duct-dependent pulmonary circulation appears to be an alternative to surgery. It provides bridging palliation until the time of definitive surgery. The effectiveness of the procedure was highlighted by the fact that all the patients showed a significant improvement in arterial saturation and pulmonary vascular marking.
Conclusion : Stenting of ductus arteriosus is feasible and safe with great result. Early detection and timely management are imperative to save the life.

Keywords : critical congenital heart defect, ductus arteriosus, ductal stenting, ductal-dependent pulmonary blood flow

INTRODUCTION

Critical congenital heart disease remains the most common cause of infant morbidity and mortality. The prevalence of congenital heart defect itself is about 6–8 in 1.000 live births, whereas 15% are life threatening defect. Congenital heart disease with duct-dependent pulmonary circulation in newborn, which identified with a severe decrease of the pulmonary blood flow, presents as a life-threatening condition. The onset of the symptom such as cyanosis or tachypnea can develop shortly after birth or within the first week of life. Typical lesions include pulmonary atresia, tricuspid atresia and Tetralogy of Fallot with pulmonary atresia.[1,2]

The presence of Patent Ductus Arteriosus (PDA) is essential for survival of newborn with duct-dependent pulmonary circulation as the source of the pulmonary blood flow. The use of Prostaglandin–E1 (PGE1) also have increased the survival of the newborn. Surgical management, however, may be limited by center experience or patient's factor such as low birth weight, prematurity and it still has high mortality risk and complication. Therefore, stenting of ductus arteriosus has been one of the management option as non-surgical approach for treating patient with duct-dependent pulmonary circulation.[3,4]

CASE REPORT

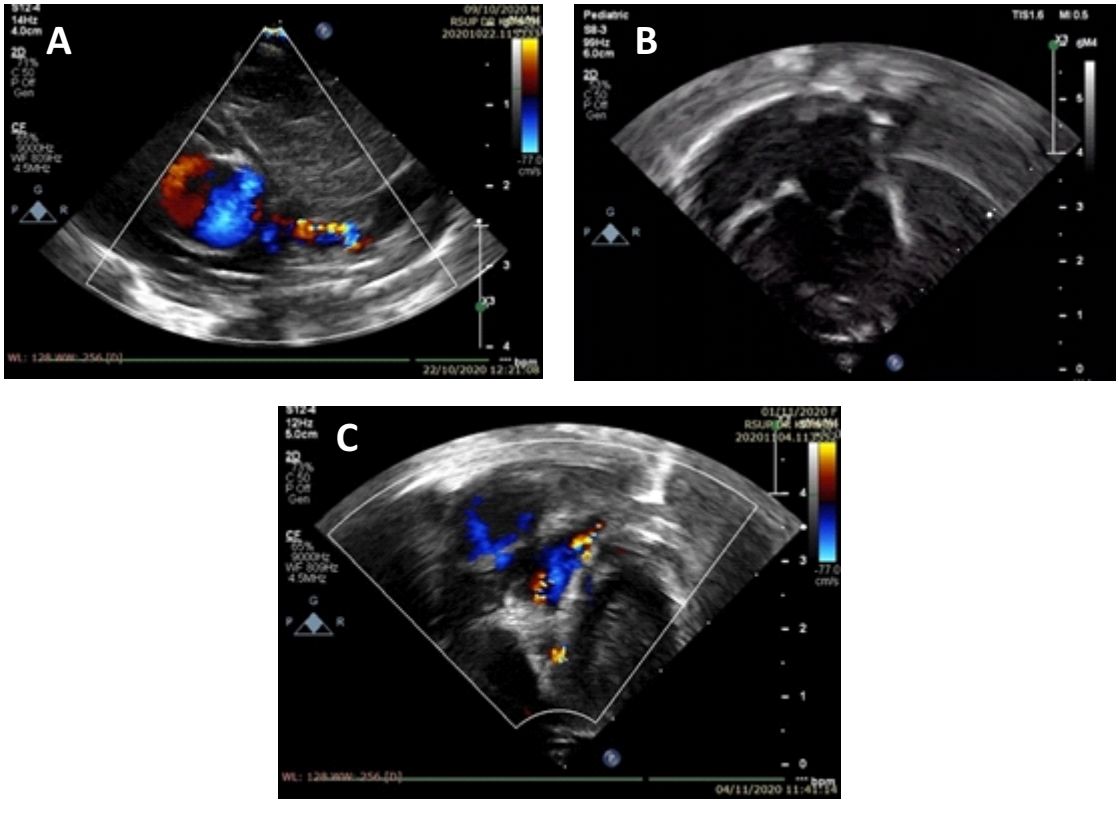
We presented newborn babies who had critical congenital heart disease with duct-dependent pulmonary circulation. All patients underwent PDA stenting. At hospital admission, patient was given PGE1 infusion until 6 hours prior to the procedure. These were the first procedure which performed in Dr. Kariadi Hospital Semarang.

Patient A

A 13-days-old male newborn with weight of 3 kg born at hospital by normal delivery was noticed to have cyanosis at 10-days-old. Oxygen saturation was 52% room air. Echocardiography showed Tetralogy of Fallot-Pulmonary Atresia (ToF-PA), restrictive Ventricular Septal Defect, and Patent Ductus Arteriosus Ø1.2–2.4 mm. Ductal stenting was performed at the age of 16 days. 1 Drug-Eluting-Stent (4.0x12 mm) was delivered in the ductal. Patient was discharged with oxygen saturation 62% room air.

Patient B

A 5-days-old female newborn with weight of 2.9 kg born at hospital by normal delivery. She was noticed to have cyanosis 2 days after birth. Oxygen saturation was 70%



Picture 1. Echocardiography on hospital admission. (A) Patient A showed Tetralogy of Fallot - Pulmonary Atresia (ToF-PA), restrictive VSD, and PDA with Ø 1.2–2.4 mm. (B) Patient B showed tricuspid atresia with restrictive VSD, atrial septal defect and PDA. (C) Patient C showed PA-IVS, Patent Foramen Ovale and PDA with Ø 1.5 mm.

room air. Echocardiography showed Tricuspid Atresia with restrictive VSD, Atrial Septal Defect (ASD) and PDA. This patient also had multiple congenital anomaly, including spina bifida, rupture of meningocele, microcephaly, and Congenital Talipes Equinovarus (CTEV). Ductal stenting was performed at the age of 8 days. 2 Drug-Eluting-Stents (3.5x9mm and 3.5x12mm) were placed in the ductal. Oxygen saturation improved until 85% room air. Patient also underwent meningocele reconstruction surgery 4 days after ductal stenting. Patient was discharged after day–12 with oxygen saturation 87% room air.

Patient C

A 2-days-old female newborn with weight of 2.2 kg born at hospital by normal delivery was noticed to have cyanosis 24 hours after birth. Oxygen saturation was 70% on CPAP. Echocardiography showed Pulmonary Atresia-Intact Ventricular Septum (PA-IVS), Patent Foramen Ovale (PFO) and Patent Ductus Arteriosus Ø 1.5 mm. Ductal stenting was performed at the age of 4 days. 1 Drug-Eluting-Stent (4.0x12mm) was delivered in the ductal. Patient was discharged after day-7 with oxygen saturation 86% room air.

The procedure was performed under general anesthesia with femoral approach. We did the aortography using 4-Fr pigtail catheter in several views to demonstrate the morphology of the ductus, measure the arterial duct accurately and administered heparin 50 IU/kg at the start of the procedures. The size of the stent was estimated according to ductal size. Drug-eluting stents was delivered by Judkins Right Guiding Catheter 3.5/5F. After stenting, aortography revealed increased pulmonary vascular, for aother marking in both pulmonary arteries and there was improvement of oxygen saturation. All patients were haemodynamically stable and then transferred to Neonatal Intensive Care Unit (NICU).

To prevent stent thrombosis, all the patients got intravenous heparin 20 IU/kg/hour for 24–72 hours post procedure and single antiplatelet with acetylsalicylic acid 5mg/kg/day. We evaluated the laboratory test, particularly the coagulation study, chest x-ray and echocardiography. Echocardiography post procedure and prior to discharge evaluation revealed the patency of ductus arteriosus. 5-month follow up after procedure, all the patients were in stable condition, and maintained with single antiplatelet. The patients were referred to prepare surgical correction.

DISCUSSION

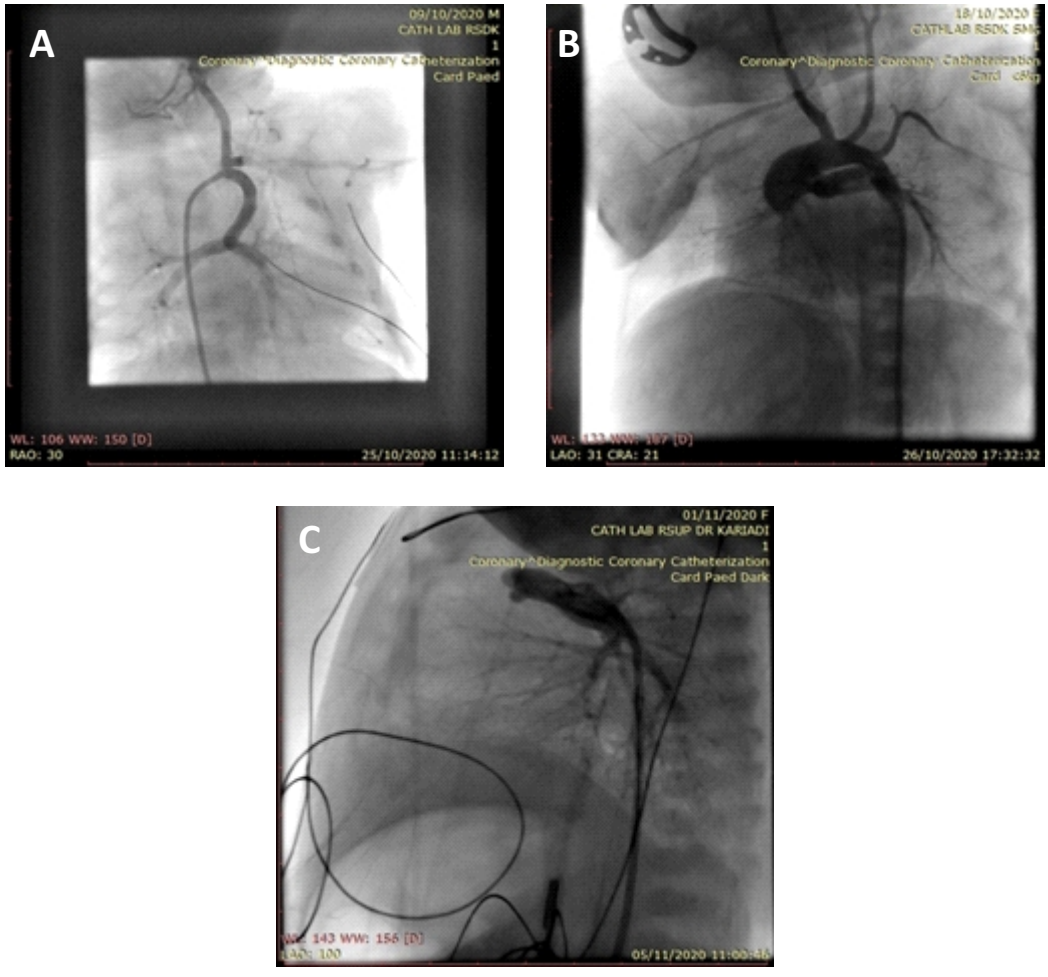
Newborns with critical congenital heart disease need early diagnosis and prompt management. Screening with pulse oximetry has been proposed as one of the strategy to establish diagnosis. Surgery remains an important first-stage palliation for critical congenital heart defect with duct-dependent pulmonary circulation. However, surgery is associated with increasing morbidity and risk of complications, especially in neonates. Therefore, ductus arteriosus stenting can be one of the management options, especially for neonates with low birth weight and prematurity, thus it will reduce the number of surgery required and optimize the time of definitive surgical correction.[3,5]

While waiting for the procedure and stabilize the critically ill neonates, we administered Prostaglandin E1 (PGE1) infusion to keep the patency of the duct. In our cases, we administered the PGE1 infusion with strict evaluation, since PGE1 might cause apnea in these small babies, and ventilatory support should be prepared. Several studies recommend to stop the infusion 6–12 hours prior to procedure. Others recommend to stop at the time of procedure when the duct had been passed by the guiding wire in severe cyanosis neonate, but we preferred to stop prostaglandin 6 hours before the procedure, to obtain the actual size of the ductus arteriosus which already started to constrict.[2,3,6,7]

The main indication for ductal stenting is to provide a bridging palliation in cyanotic congenital heart disease before performing definitive surgery. According to American Heart Association, stenting of Patent Ductus Arteriosus as the source of pulmonary circulation in an infant with cyanotic congenital heart disease, is classified as class IIB indication. In our cases, we had neonates with

TABLE 1
Details of all the patient including complication

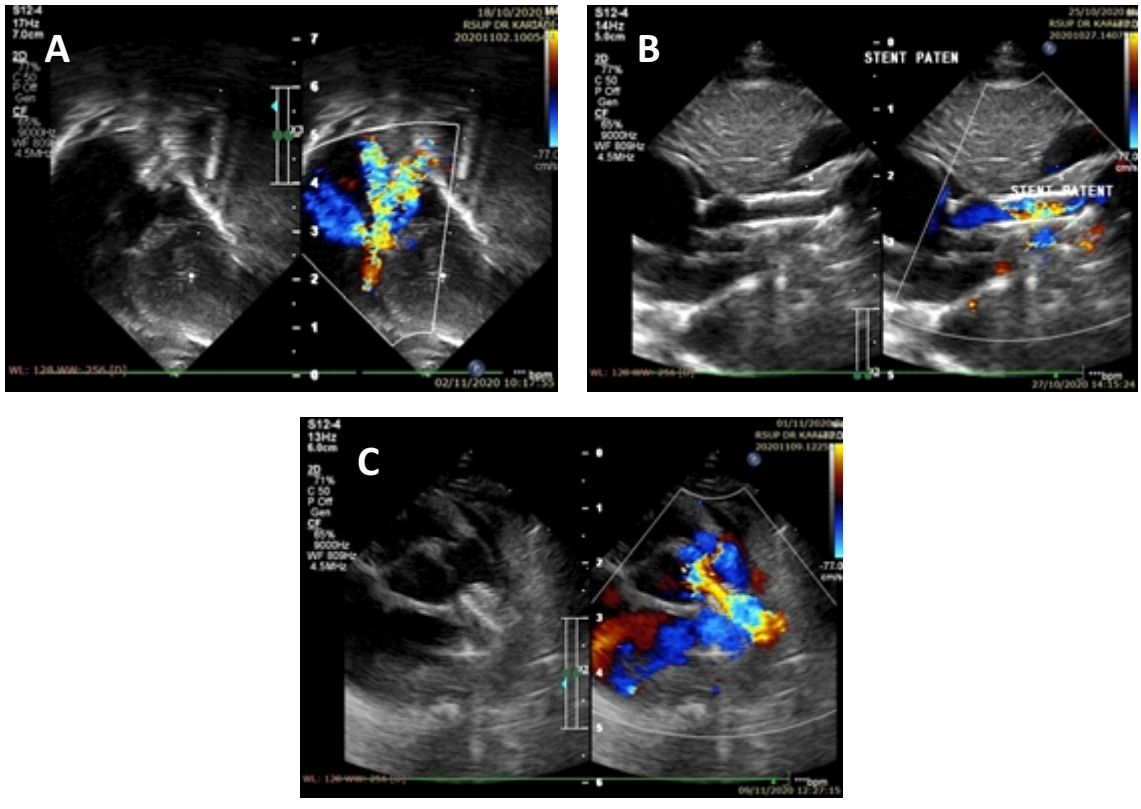
Patient	Age/Sex	Weight (gram)	Diagnosis	Stent size (mm)	Complication	Death before discharge
A	13 days/M	3000	ToF-PA	4.0x12	No	No
B	5 days/F	2935	Tricuspid atresia, restrictive VSD	3.5x9, 3.5x12	No	No
C	2 days/F	2200	PA-IVS	4.0x12	No	No



Picture 2. Angiogram showed contrast flow in PDA and pulmonary arteries after stenting in patient A, B, and C

tetralogy of fallot with pulmonary atresia (ToF-PA), pulmonary atresia with intact ventricular septum (PA-IVS) and tricuspid atresia with restrictive ventricular septal defect, whereby all of them depended on ductus arteriosus for the pulmonary circulation. Ductal stenting should not be performed in case of branch pulmonary artery stenosis.[5,8] Prior to the procedure, we obtained detailed echocardiography to evaluate the structure and anatomy. It was essential to recognize the morphology of the PDA and pulmonary artery. The procedures were performed in the catheterization laboratory with general anesthesia and femoral artery approach. Aortic angiography was performed to assess the PDA, pulmonary artery, and aortic arch. Horizontal and tubular duct has a higher success rate for stenting. Heparin injection with body weight adjusted dose was administered to achieve an activated clotting time >250 seconds. Afterwards, 5F Judkins Right guiding catheter which able to support delivery of the stent, was engaged in PDA ampulla, and a 0.014–inch coronary wire was gently crossed over the PDA and anchored in one of distal pulmonary artery branch. Once the stent position was obtained, the stent was inflated according to the pressure required. Repeated angiography was done to evaluate the stent position, pulmonary circulation, pulmonary vascular marking and determine whether additional stent is required. We successfully delivered drug eluting stent (DES) in all of the patients. Aortography post stenting revealed increased pulmonary vascular marking in the patients. Study reported that using drug eluting stent resulted in less luminal loss of ductal arteriosus and decrease unplanned reintervention as compared with bare metal stent implantation in infant with ductal dependent pulmonary circulation.[5,8,9]

Following the procedure, the patients were transferred to Neonatal Intensive Care Unit (NICU) for hemodynamic monitoring. We administered heparin infusion for 2–3 days post procedure and started single antiplatelet acetylsalicylic acid 5mg/kg/day to prevent stent thrombosis. There was lack of study regarding to the use of dual antiplatelet therapy with addition of



Picture 3. Echocardiography prior to discharge showed patency of ductus arteriosus

clopidogrel for neonates who undergo ductal stenting. Oxygen saturations were improved in all of the patients and there was no post-procedural complication reported. Echocardiography which performed after the procedure and prior to discharge showed stent patency and flow in the ductus arteriosus.[6, 8, 10, 11]

The majority of patients underwent PDA stenting at an early age, usually shortly after birth, with weight ranging between 2.2–3.0 kg. Stent placement was successful in all of the cases. The effectiveness of the procedure was highlighted by the fact that all the patients showed a significant improvement in pulmonary vascular markings and arterial saturation after stenting. Ductal stenting is now preferred as an alternative to surgery in patient with duct-dependent pulmonary circulation. A retrospective cohort study by Glatz e*t al* compared PDA stent and modified Blalock-Taussig Shunt as palliation in 357 infants with ductal-dependent pulmonary blood flow. The study showed no difference in mortality and unplanned reintervention to treat cyanosis between two groups. However, PDA stent group showed shorter length of stay in intensive cardiac care unit, decrease the risk of diuretic use at discharge, and larger and more symmetrical pulmonary arteries at the time of surgical repair. Therefore, this study supported PDA stent as an alternative to Blalock-Taussig Shunt in infants with ductal-dependent pulmonary circulation. Several studies also showed that ductal stenting was effective and feasible comparing to surgery.[12-15]

CONCLUSION

Our cases demonstrated cyanotic congenital heart defect with ductal-dependent pulmonary circulation successfully managed by ductus arteriosus stenting. Ductal stenting can be an alternative to surgery to decrease morbidity and mortality in newborn with duct-dependent pulmonary circulation. Applying coronary drug eluting stent has made this procedure feasible with great result. Early detection and timely management are imperative to save the life.

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