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First Experience of Patent Ductus Arteriosus Stenting in Neonate with Pulmonary Atresia and Intact Ventricular Septum in North Sulawesi, Indonesia

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Abstract: Stenting of patent ductus arteriosus (PDA) is a minimally invasive catheter-based temporary palliative procedure, and it serves as an alternative to surgical shunt in neonates with duct dependent pulmonary perfusion, such as pulmonary atresia. This presentation aimed to report the first experience of PDA stenting in a neonate with pulmonary atresia in North Sulawesi, Indonesia. We reported a female neonate patient, weighing 3775 grams, referred from primary hospital with suspect congenital heart disease. The patient was born at 40 weeks of gestational age by caesarean section delivery from a primigravid mother aged 23 years with inertia uteri and strangulation of nuchal cord. At birth, she cried immediately and her APGAR score was 6-7. On her first day of life, she looked cyanotic, therefore she was referred to Prof. Dr. R. D. Kandou Hospital. Physical examination showed that she had perioral cyanotic and peripheral cyanotic with SpO2 58%. Chest X-ray showed cardiomegaly. Echocardiography revealed pulmonary atresia with PDA with intact ventricular septum. PDA stenting was done on her second day of admission. After the procedure, she was clinically stable with SpO2 80%, and was treated with intravenous antibiotic, and intravenous heparin and aspirin enterally. On the third day post procedure, she was discharged and no complication was found. PDA stenting in patient with duct-dependent pulmonary circulation appeared to be an alternative to surgery. It provided bridging palliation until the time of definitive surgery. The effectiveness of the procedure was highlighted by the fact that all the patients showed significant improvement in arterial saturation and pulmonary vascular marking. In conclusion, PDA stenting is considered feasible, safe and associated with lower mortality rates and shorter hospital length of stay than systemic-pulmonary surgical shunt procedure. Early detection and timely management are imperative to save the life.
Keywords: patent ductus arteriosus stenting; pulmonary atresia

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% of them are life threatening defects. Congenital heart disease with duct-dependent pulmonary circulation in newborn 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]

In pulmonary atresia, there is no communication between the right ventricle and the pulmonary artery, therefore, a patent ductus arteriosus (PDA) or no collateral arteries become a major source of blood flow to the lungs. Neonates with ductal dependent pulmonary blood flow would benefit from maintained ductal patency for several months until he/she is ready to undergo a definitive surgery. Conventional palliative therapy to maintain pulmonary blood flow is to provide a prostaglandin-E1 (PGE1) infusion. Surgical management, however, may be limited by center experience or patient's factor such as low birth weight or prematurity, and it still has high mortality risk and complication. Therefore, stenting of ductus arteriosus has been one of the management option as a minimally invasive catheter-based temporary palliative procedure, which serves as an alternative to surgical shunt for treating patient with duct-dependent pulmonary circulation. In this study, we reported a case of pulmonary atresia with successful PDA stenting as an alternative to maintain pulmonary blood flow.[2]

CASE REPORT

A female neonate, weighing 3775 grams, was referred from primary hospital with suspected congenital heart disease. She was born at 40 weeks of gestational age by caesarean section delivery from a primigravid mother aged 23 years with inertia uteri and strangulation of nuchal cord. At birth, she cried immediately and her APGAR score was 6-7. On her first day of life, she looked cyanotic, therefore she was referred to Prof. Dr. R. D. Kandou hospital. In physical examination, she had perioral cyanotic and peripheral cyanotic with SpO2 58%. Chest X-ray showed cardiomegaly. Echocardiography showed pulmonary atresia with a PDA and intact ventricular septum. PDA stenting was done on her second day of admission. After the procedure, she was clinically stable with SpO2 80%. She was treated with intravenous antibiotic, intravenous heparin, and acetylsalicylic acid enterally. On the third day post procedure, she was discharged and no complication was found.

DISCUSSION

Newborns with critical congenital heart disease need early diagnosis and prompt management. Screening with pulse oximetry has been proposed as one of the strategies to establish the 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 surgeries required and optimize the time of definitive surgical correction.[1,2] Ductal stenting primary indication is when neonates are at high risk for surgery, unavailability of adequate surgical facilities, ideal ductal morphology (tubular/ straight duct), and the need to maintain ductal patency for more than 2 weeks.

While waiting for the procedure and stabilizing the critically ill neonates, prostaglandin E1 (PGE1) infusion is administered to keep the patency of the duct. Several studies recommend to stop the infusion 6–12 hours prior to the procedure. Others recommend to stop at the time of procedure when the duct has been passed by the guiding wire in severe cyanosis neonates, but it is preferred to stop prostaglandin 6 hours before the procedure, to obtain the actual size of the ductus arteriosus

which already started to constrict.[1,3-54]In our case, PGE-1 infusion was not available.

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 PDA as the source of pulmonary circulation in an infant with cyanotic congenital heart disease, is classified as class IIB indication. In our case, we had a neonate with pulmonary atresia with intact ventricular septum (PA-IVS).

Prior to the procedure, we performed a detailed echocardiography to evaluate the structure and anatomy of cardiac blood vessels. It was essential to recognize the morphology of the PDA and pulmonary arteries. 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. Oxygen saturation was improved in the patient and there was no post-procedural complication reported.

Following the procedure, the patient was 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. The effectiveness of the procedure was highlighted by the fact that the patient showed a significant improvement in oxygen saturation after stenting.

Ductal stenting is now preferred as an alternative to surgery in patients with duct-dependent pulmonary circulation. A retrospective cohort study by Glatz et 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 the 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.[6-9,] Ductal stenting could reduce hospital length of stay, the infants could be discharged in 48 hours after the procedure. Evaluation is usually done 6-12 weeks after that until definitive surgery performed.[4] Moreover, surgery after stenting is safe and has a low risk.[10]

CONCLUSION

In neonates with ductal-dependent pulmonary circulation, such as pulmonary atresia, PDA stenting is one of the alternative treatments to the classical surgical shunt procedure. This procedure is considered feasible and safe and is associated with lower mortality rates and shorter hospital length of stay than systemic-pulmonary surgical shunt procedures.

Conflict of Interest

The authors affirm no conflict of interest in this study.

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