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Case Report

The Success of an Intra-Ductal Stenting in a Three-Day-Old Infant with Tricuspid Valve Atresia: A Case Report

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ABSTRACT

Tricuspid valve atresia is a congenital heart defect and if left untreated, the lesion has a high mortality rate. This study aimed to report a case of tricuspid valve atresia in a low-birth-weight three-day-old infant, who was treated by intra-ductal stenting and established pulmonary blood flow instead of routine shunting.

The patient was a three-day-old male infant, weighing 2800 grams with a gestational age of 38 weeks. He was admitted with symptoms of tachypnea, cyanosis, 45% saturation, and severe metabolic and respiratory acidosis. On echocardiography, tricuspid valve atresia was observed. The infant was treated with prostaglandin E1 immediately after admission to the NICU and necessary interventions were taken to keep the patent ductus arteriosus (PDA) open. Angiography was performed through the aortic artery. First, a balloon was inserted, and then a stent was successfully placed inside the PDA. Preoperative saturation increased from 45% to 93% after stenting, and the infant was extubated after 48 hours in a good general condition.

As the results of the current case report showed, stent placement can effectively keep the PDA open and establish pulmonary blood flow in high-risk infants. It is suggested that ductal stenting should be considered as the first treatment selection in newborns with tricuspid valve atresia or as a good alternative method for Blalock-Taussig shunt.

Keywords: Arterial duct stent, Infant, Tricuspid atresia, Congenital defect, Prostaglandin

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Introduction

There are different types of tricuspid valve atresia (TVA) with different clinical manifestations based on the amount of the pulmonary blood flow, which will cause a very high mortality rate if left untreated. Tricuspid valve atresia comprises about one percent of congenital heart diseases, and its incidence rate is one per 10,000 cases, being equal between men and women (1). Approximately, 50% and 80% of infants with this disease show symptoms on the first day of birth and during the first month of birth, respectively (2). In infants with pulmonary artery obstruction, systemic arterial oxygen saturation decreases. As a result, these infants are usually cyanotic with a high probability of clot formation, heart attacks, and stroke. A fully systolic heart murmur in the left lower sternal border is a sign of a ventricular septal defect. In addition, a systolic ejection murmur may be heard in the left lower sternal border. Prostaglandin E1 is the primary treatment for maintaining pulmonary circulation in infants with TVA, if it is not taken, closure of the ductus arteriosus occurs within two to three days after birth (3). Surgical treatments in these patients include a balloon atrial septostomy, a modified BT shunt between the aortic artery and the pulmonary artery, pulmonary artery bands, the Fontan method, and the Glenn procedure for connecting the superior vena cava to the pulmonary artery (4). This study was conducted to report a TVA case in a three-day-old infant who, despite his young age, was treated using a PDA stent instead of routine shunting treatment.

This case report was approved by the ethics committee of Kerman University of Medical Sciences (code: IR.RUMS.REC.1399.188).

Characteristics of the Case

The patient was a three-day-old boy, born via a natural vaginal delivery, term, weighted 2,800 g, with no congenital and familial medical history, who was dispatched to Afzalipour hospital in Kerman with symptoms of tachypnea, cyanosis, 45% saturation, and severe metabolic and respiratory acidosis. On clinical examination load S2 (the second heart sound) and the systolic murmur were heard in LSB (left sternal border). Initial echocardiography showed TVA. The infant could survive with a single ventricle defect and with the open typical tubular PDA. So the patient was treated with prostaglandin E1 (0.1 mg/kg/min) immediately after admitting to the NICU (neonatal intensive care unit) to keep the PDA open and received the necessary treatment (300 CC dextrose water 10% to keep the urine specify gravity level between 1010 to 1015) and respiratory support (PEEP= 3, PIP=18, IT=0.5, Sat O2= 45% and RR=45). When saturation reached an acceptable level (75%), we gradually reduced the prostaglandin dose to 0.01 mg/kg/min. Due to the patient's young age and the high risk of cardiac arrest, sepsis, and mortality, it was decided to place a stent inside the PDA through angiography. During angiography, we entered the aortic artery and pre-dilated with the balloon (Mozec 4*13) inflated and the stent (coroner lineage 4*16) having been successfully placed inside the PDA. Figure 1 shows the stent placement steps. Accordingly, oxygen saturation increased from 75% (before surgery) to 93% (after surgery), and the infant was extubated after 48 hours under good conditions. He was discharged from the cardiac ward, but the neonatal ward preferred a full course (seven days) of antibiotic treatment. After surgery echocardiography showed continuous flow from the aorta to the pulmonary artery. In the follow-up period, he was readmitted after 20 days due to overflow, treated with diuretics, and was discharged again with proper feeding and good general condition. After a month, he had good feeding and gained proper weight. The next plan for this patient at the age of six to eight months is to perform a Glenn operation and then a Fontan procedure after the baby starts walking.

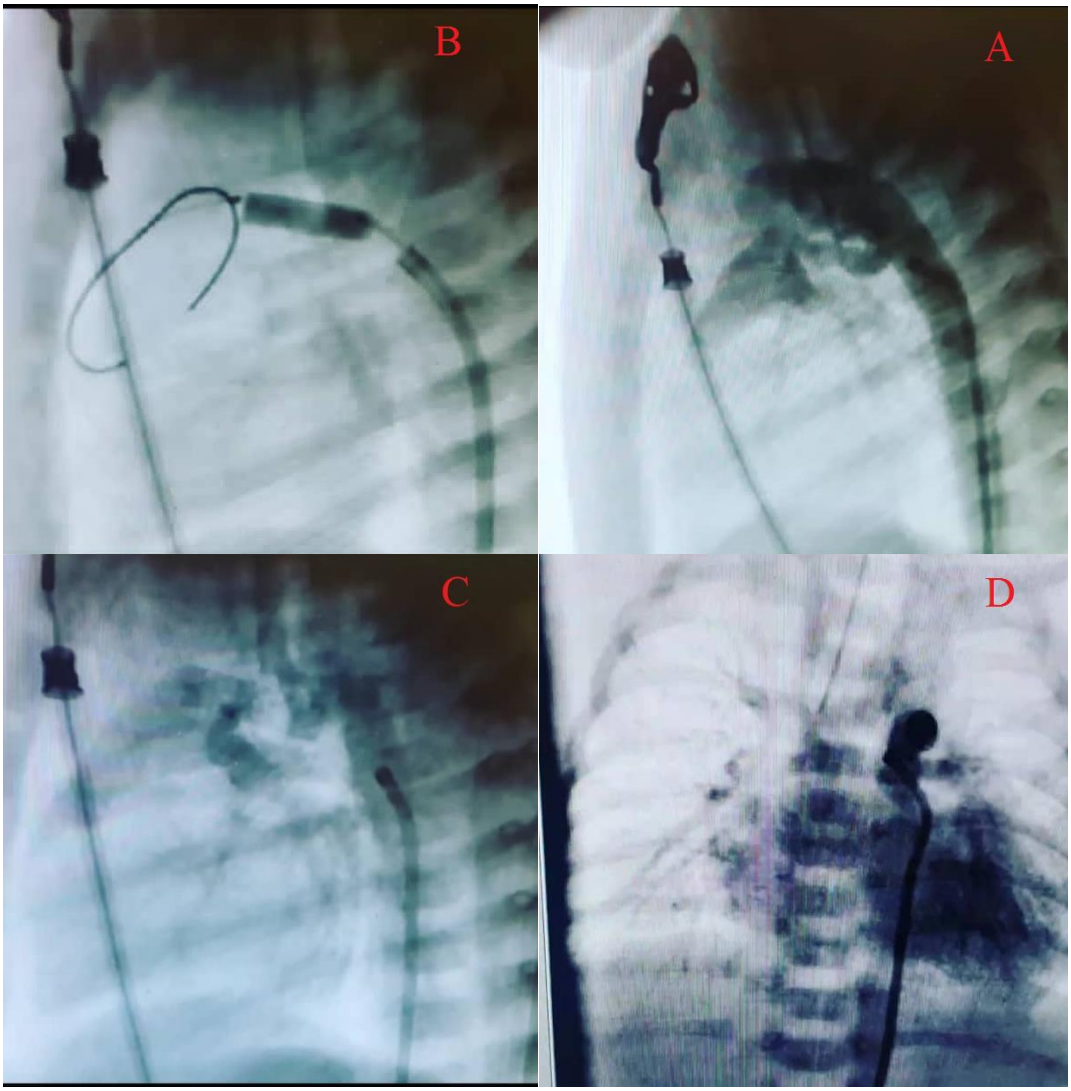

Figure 1. Stages of stent placement inside PDA through angiography in an infant with TVA

(A. The lateral view of aortic arch injection: the tubular PDA with multiple stenoses connects the aortic artery to the pulmonary artery; B. The lateral view of aortic arch injection: the PDA dilatation stage with a coronary balloon; C. The lateral view of aortic arch injection: stages of coronary stent placement in the PDA; D. The frontal view of injection in the pulmonary artery after stent placement: filling both pulmonary arteries equally)

Discussion

Tricuspid valve atresia is a congenital heart defect occurring along with other heart defects in infants, which requires various treatment stages, and if left untreated, it will be fatal (2). The results of a review study showed a lack of evidence-based education for patient identification, evaluation, management, and treatment, as well as permanent consequence identification in these patients (5). As the results of the present study showed, after prostaglandin E1 infusion to impending ductal closure, the intra-ductal stent was successfully placed inside the PDA. In the same study done by Kumar *et al.*, a newborn baby who had TVA and hypoplastic right ventricle was successfully carried out with ductal stenting (6). In a single-centre experience report by Odemis *et al.*, while 13 patients underwent PDA stenting, five deaths were observed due to procedure-related conditions, pulmonary hemorrhage, retroperitoneal hemorrhage, sepsis, and aspiration pneumonia. Eight of thirteen patients achieved ductal stent effectively (7). Early diagnosis and timely interventions are very important for saving these infants' lives. Surgery and shunting are common treatments for congenital neonatal heart disease, which have been validated in treating TVA. However, these methods increase the mortality rate of infants, especially those weighing less than 3000 g (8). Stent placement is another way of treating this disorder. Research shows that the use of this method improves the growth of pulmonary

arteries and reduces complications and hospital stay, yet it increases neonatal survival (9). Stent placement can effectively keep the PDA open and increase the pulmonary blood flow in high-risk infants. The new generation of coronary stents has better properties, flexibility, and traceability, and is placed more easily. Hence, ductal stenting is a suitable alternative treatment for shunt surgery in neonates with ductal-dependent congenital heart defects, such as TVA, because it prevents thoracotomy and the related adverse complications (10). Therefore, the treatment complications and duration could be reduced in patients with ductal-dependent cyanotic congenital heart defects by selecting appropriate conditions, patients, techniques, and tools.

Conclusion

Timely identification and treatment of high-risk infants with TVA are the first steps in saving their lives. Stent placement could effectively keep the PDA open and maintain the pulmonary blood flow in infants; in addition, it reduces complications, obviates surgical procedures, reduces the admission duration, and increases neonatal survival. It is suggested that ductal stenting should be considered as a first treatment selection in newborns with TVA or as a good alternative method for BT shunting.

Clinical Application of Findings

The results of this study indicate that this method reduces complications and increases the survival rate in high-risk infants.

References

1. Alahmadi DH, Alharthi WM, Jifri AT, Alkhabbaz MJ, Alharbi MK, Alruwaili HI, *et al.* Evaluation of recent updates regarding the diagnosis and management of congenital heart in children. Arch Pharma Pract. 2019; 10(3):29-32.
2. Sumal AS, Kyriacou H, Mostafa AMHAM. Tricuspid atresia: Where are we now? J Card Surg. 2020; 35(7):1609-17. doi: 10.1111/jocs.14673.
3. Akkinapally S, Hundalani SG, Kulkarni M, Fernandes CJ, Cabrera AG, Shivanna B, *et al.* Prostaglandin E1 for maintaining ductal patency in neonates with ductal-dependent cardiac lesions. Cochrane Database Syst Rev. 2018; 2(2):CD011417. doi: 10.1002/14651858.CD011417.
4. Park IS, Kim SJ, Goo HW. Tricuspid atresia. In: Park, I. (ed). An illustrated guide to congenital heart disease. Singapore: Springer; 2019. p. 253-67. doi: 10.1007/978-981-13-6978-0_12.
5. Frock BW, Jnah AJ, Newberry DM. Living with tricuspid atresia: case report with review of literature. Neonatal Netw. 2017; 36(4):218-28. doi: 10.1891/0730-0832.36.4.218.
6. Kumar P, Datta R, Nair R, Sridhar G. Stent implantation of patent ductus arteriosus in a newborn baby. Med J Armed Forces India. 2011; 67(2):171-3. doi: 10.1016/S0377-1237(11)60027-0.
7. Odemis E, Haydin S, Guzeltas A, Ozyilmaz I, Bilici M, Bakir İ. Stent implantation in the arterial duct of the newborn with duct-dependent pulmonary circulation: single centre experience from Turkey. Eur J Cardiothorac Surg. 2012; 42(1):57-60. doi: 10.1093/ejcts/ezr258.
8. 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):642-51. doi: 10.1016/j.athoracsur.2011.02.030.
9. 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. Congenit Heart Dis. 2019; 14(1):105-9. doi: 10.1111/chd.12707.
10. Alwi M. Stenting the ductus arteriosus: Case selection, technique and possible complications. Ann Pediatr Cardiol. 2008; 1(1):38-45. doi: 10.4103/0974-2069.41054.