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CLINICAL CASE

Stent ductal implant by axillary via in a newborn with ductal dependent pulmonary blood flow

Implante de *stent* ductal por vía axilar en neonato con cardiopatía congénita con flujo pulmonar dependiente del conducto arterioso

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What do we know about the subject matter of this study?

Ductal stenting in arterial ducts with unusual anatomies and origins represents a challenge for pediatric cardiac intervention. The axillary artery route offers a safe and effective option for access to these ducts.

What does this study contribute to what is already known?

This case report shows the feasibility of using arterial routes other than the femoral one in low-weight patients requiring ductal stenting in duct-dependent pulmonary circulation.

Abstract

Ductus arteriosus stenting is a palliative alternative for neonates with ductal-dependent pulmonary flow. **Objective:** To present an alternative of arterial access for percutaneous coronary intervention in neonates. **Clinical Case:** A term neonate with low weight diagnosed with pulmonary atresia with intact ventricular septum and severe hypoplasia of the tricuspid valve with dependent coronary circulation. Due to the surgical risk and femoral artery damage and the anatomy of the ductus arteriosus, a left axillary arterial puncture was decided where a 3.5-millimeter coronary *stent* was successfully placed. The patient developed an axillary spasm that resolved spontaneously. **Conclusion:** Alternative arterial access other than the femoral artery route is an option for neonates with high surgical risk and low birth weight.

Keywords:
Congenital Heart Defects;
Cardiac Catheterization;
Stents;
Therapeutic Use;
Complications

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Introduction

Stent implantation in the ductus arteriosus (DA) has been shown as a palliative alternative to Blalock-Taussig shunt in newborns (NBs) with obstructive lesions in the right ventricular outflow tract[I,II]. This procedure allows safe pulmonary flow while performing the surgical correction of the underlying heart disease and promotes the growth of the pulmonary arterial tree which results in a better balance of the pulmonary vasculature, thus the pulmonary branches grow with more symmetry[III,IV,V]. However, variations in the origin, orientation, and morphology of the DA in those congenital heart diseases where pulmonary flow is compromised, require a change in the usual approach through the femoral artery due to the difficulty of this access when the ductal angle is greater than 90º. The tortuous nature of the ductus arteriosus in these patients, and patients under 15 kilograms, presents an exacerbated difficulty. Femoral access has been associated with a higher risk of thrombosis[VI,7].

Other vascular accesses that have been used include the femoral vein, however, it is not an option for all types of heart disease and the carotid artery is another effective option for stenting in NBs with good alignment of the conduit[VII,8]. Another approach is through the axillary artery, which has shown few complications and, similarly to the carotid access, allows direct duct alignment, faster access, and a greater chance of success than access through the femoral artery[VIII].

The objective of this case report was to present the percutaneous management of an NB with pulmonary atresia who underwent DA stenting via the axillary route. Informed consent was obtained and information confidentiality was guaranteed. The international ethical guidelines for health-related research involving humans by the Council for International Organizations of Medical Sciences (CIOMS) were followed. This paper was prepared following the CARE guidelines for case reports[IX].

Clinical Case

Male newborn of 37+1 weeks of gestational age with prenatal diagnosis of intrauterine growth restriction type I. Spontaneous vertex delivery, cephalic presentation, APGAR score 6 at one minute and 8 at five minutes. Weight 2,180 g, length 45 cm. Since the patient presented poor neonatal adaptation, he required continuous positive airway pressure (CPAP) due to increased work of breathing and hypoxemia. Due to respiratory deterioration, he was transferred to neonatal intensive care, performing orotracheal intubation and start administration of prostaglandin E1. The first echocardiography showed pulmonary atresia with intact ventricular septum; 78% oxygen saturation, FiO2 50%, heart rate 165 beats per minute (bpm), blood pressure 80/32/48mmHg, rhythmic heart sounds with a single second sound. On the second day of life, the NB was referred to a fourth-level cardiovascular center for congenital heart disease (figure 1).

Diagnostic Aids

Echocardiography showed pulmonary atresia with intact ventricular septum, tricuspid valve with severe hypoplasia (Z score-4.04), and abnormal flows compatible with intramyocardial sinusoids which leads to the suspicion of right ventricle-dependent coronary circulation; restrictive patent DA with left-to-right shunt, having a tortuous form with at least two curves. No origin of the lesser curvature of the aortic arch is observed. Confluent pulmonary branches and mild stenosis at the beginning of the left branch. Non-restrictive patent foramen ovale with a right-to-left shunt; severe right ventricular hypertrophy and hypoplasia; bipartite right ventricle; right atrium dilatation.

The CT scan showed confluent pulmonary arteries, right pulmonary artery of 2.5 mm and the left one of 1.8 mm, patent DA from the left lateral aspect of the aortic arch with anterior and medial direction towards the dorsal aspect of the proximal left pulmonary artery. From the aorta, the ductus measures 2.5 mm at the beginning, 1.5 mm in the middle third, and 2 mm at the end in the left pulmonary artery. Low-attenuation material towards the tricuspid and pulmonary valve compatible with pulmonary atresia and tricuspid hypoplasia. Bovine aortic arch. Normal systemic and pulmonary venous connections. Significantly decreased left main bronchus 4 mm next to the carina, predominantly in the anteroposterior plane (1 mm vs 4 mm of the contralateral) due to hypoplasia or bronchomalacia. A 3D reconstruction was performed, and it was decided to approach through the left axillary artery (figure 2).

On the fourth day of life of the patient, the case was discussed at a medical-surgical meeting, with the participation of pediatric cardiologists (echocardiographers, hemodynamics specialists, intensivists), pediatric heart surgeons, and pediatric cardiovascular anesthesiologists. Since this was a low-birth-weight NB with a DA of anomalous origin, it was decided to perform cardiac catheterization to evaluate the coronary circulation and the possibility of implanting a ductal stent. It was decided to use the axillary approach because it was thought to be the most direct route for stent implantation, although the possibility of a transcarotid approach was discussed, which is another option for this type of complex anatomy.

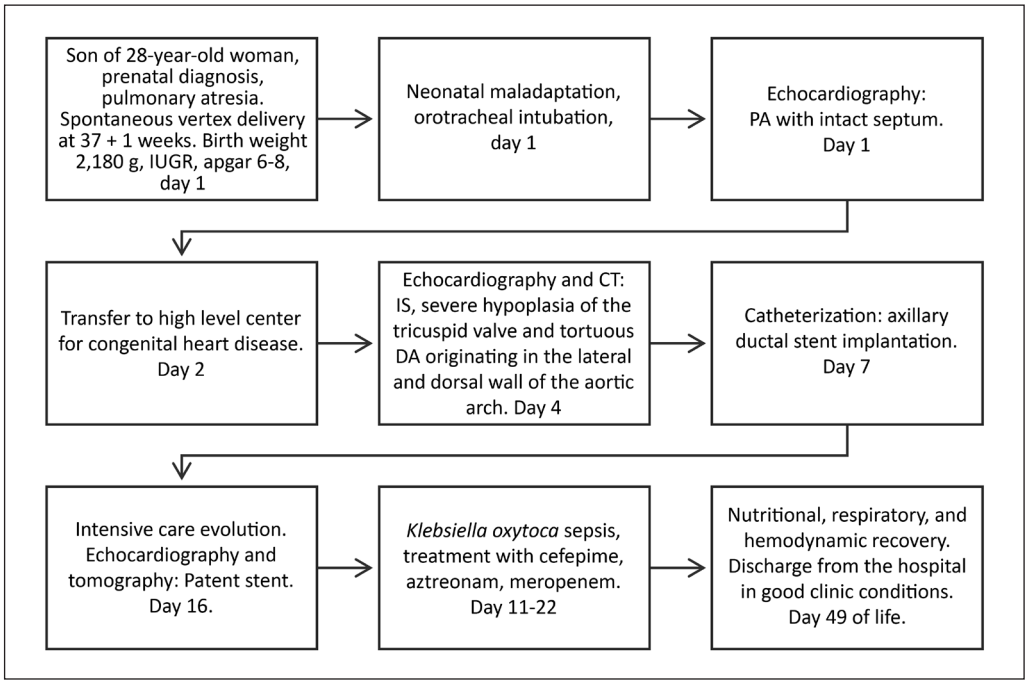

Figure 1. Timeline. IUGR: Intrauterine growth restriction; PA: Pulmonary atresia; CT: Computed tomography; IS: Intact septum; DA: Ductus arteriosus.

Cardiac Catheterization

At seven days of life, he is taken to the cardiac catheterization room using a biplane x-ray system (Allura Xper®, Phillips, Holland). The patient is placed upside down (feet on top of the table) which allows hemodynamics specialists to access safely and comfortably the left axillary artery, in addition to an adequate visualization of the angiography monitors. The procedure was performed with 100% oxygen, orotracheal intubation, and cardiovascular anesthesia. Informed consent was obtained and due to the COVID-19 pandemic, full personal protection measures were taken.

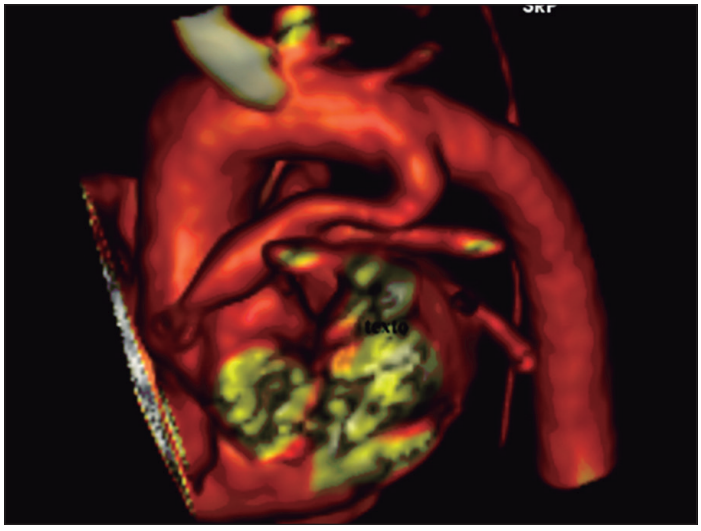

Figure 2. 3D Volumetric representation of the aortic arch with tortuous ductus arteriosus configuration.

According to the institutional protocol in patients with DA-dependent pulmonary circulation, the administration of prostaglandin E1 was suspended 1 hour before starting the procedure.

Description of the Procedure

Under vascular ultrasound guidance, direct puncture of the left axillary artery was performed with needle No. 21. When blood reflux was obtained through the needle, a 0.18" micropuncture guide was introduced, the needle was withdrawn and a 4 French (F) Terumo® introducer was inserted over the guide. A Non-Taper Angle® catheter (Glidecath Radiofocus, Tokyo), 4F on a 0.035" hydrophilic guide was inserted through this introducer. Angiography with manual injection showed bilateral coronary involvement. The right coronary artery presented distal atresia and only its proximal region was identified and in the left coronary artery, areas of severe stenosis were observed in the anterior descending artery. Therefore, it was considered that the coronary circulation was dependent on the right ventricle as it did not present adequate antegrade filling. A thoracic aortogram was performed to evaluate the dimensions of the DA which showed pulmonary end 1.5 mm, aortic end 3 mm, and length of 8 mm. The Balance Heavy Weight® coronary guide wire (Abbott, United States), 0.014", advanced with difficulty to the pulmonary branches due to the areas

of stenosis and the anomalous origin of the DA in the aorta. Due to the difficulty in advancing the guide wire because of the tortuous form of the DA, a distal microcatheter was introduced in the right branch, advancing the guide wire deeper. The guide wire was positioned distally in the right branch of the pulmonary artery and later a 0.014" distal guide wire was advanced in the left branch to perform the buddy wire technique (use two simultaneous guide wires to facilitate the advancement of the stent in the DA), which continued without complications.

A 3.5 mm x 12 mm medicated stent (Medtronic®, Ireland) was placed in the DA with a pressure of 10 atm. The stent diameter was selected according to the patient's weight and length based on angiographic and tomographic measurements, in order to completely cover the whole DA to avoid leaving areas of the DA not covered by the stent.

Control angiography showed the stent in good position, with complete filling of the pulmonary branches without extravasation of contrast medium and axillary artery spasm. There were no other complications. Fluoroscopy time was 49 minutes, catheterization procedure lasted 120 minutes, and 25 ml of non-ionic iodinated contrast media was used. (figures 3 and 4).

Intensive Care

Once the catheterization was completed, the patient returned to the intensive care unit for ventilatory support and comprehensive care. Heparin was administered IV at 28 units/Kg/hour, to avoid acute stent thrombosis and management of arterial spasm with partial thromboplastin time control and subsequent change to antiplatelet treatment with aspirin and clopidogrel. The following day the spasm was resolved, and the patient presented radial pulse.

One of the pathophysiological events that can occur in this type of patient is pulmonary hyperflow that can trigger congestive heart failure that is difficult to control. This pathophysiological event depends largely on the size of the implanted stent. Another frequent complication in this type of patient is the "ductal steal", which occurs when the diastolic blood pressure falls, leading to coronary insufficiency and ischemia. Neither of these two complications occurred in our patient.

During the evolution, the NB presented episodes of oxygen desaturation and hypoxemia, therefore an echocardiogram and tomography were requested, which showed stent permeability (figure 5). Subsequently, sepsis due to *Klebsiella oxytoca* (R) was diagnosed, which was treated with antibiotics resulting in negative blood cultures. After 22 days of hospitalization in the ICU, he was transferred to pediatric cardiology where he remained for another 27 days until the sepsis and enteral feeding problems were resolved and the patient's evolution and the stent were closely monitored.

The 49 days of hospitalization can be explained not only by the complexity of the heart disease, and the risks inherent to the intensive care stay such as physical deconditioning, loss of enteral nutrition, use of central catheters, and sepsis, but also by the socio-familial circumstances.

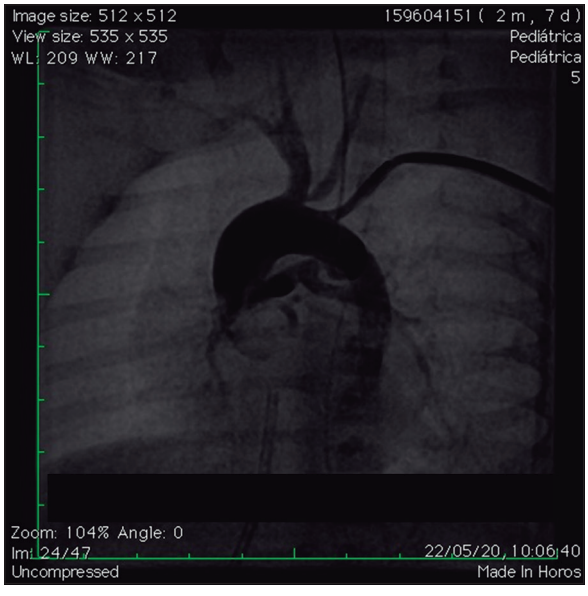

Figure 3. Angiography via the left axillary artery, the tortuous ductus arteriosus originating from the lower region of the aortic arch is observed.

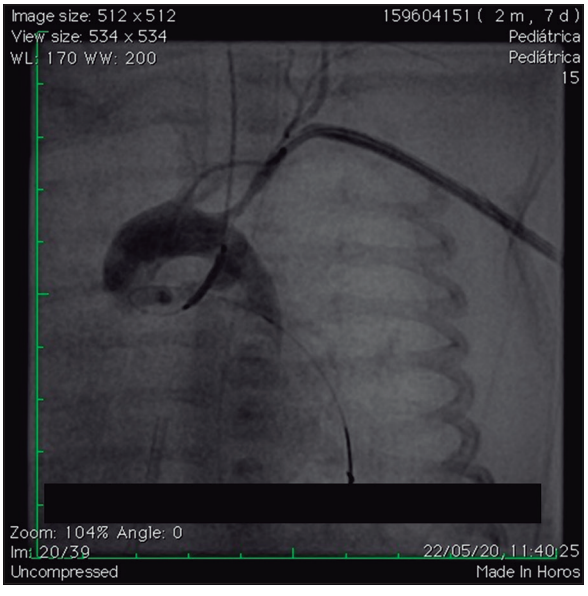

Figure 4. Angiography via the left axillary artery, patent ductus arteriosus with stent in place is observed.

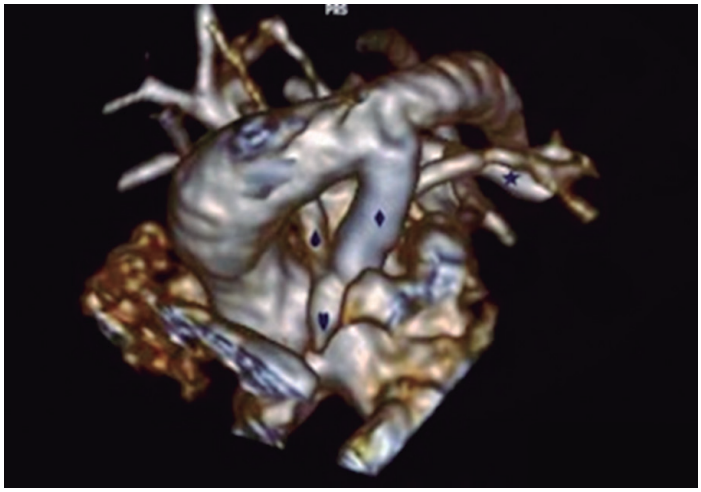

Figure 5. 3D volumetric representation of ductal stent.

Discussion

We describe the implantation of a medicated ductal stent via the axillary route in a 7-day-old low-birthweight NB with DA-dependent pulmonary circulation secondary to pulmonary atresia with intact ventricular septum, severe tricuspid valve hypoplasia, and right ventricle-dependent coronary circulation.

In 2004, Michel-Behnke et al published the first reports of this access route for neonatal intervention and subsequently published other case series with emphasis on the technique. To our knowledge, few cases of axillary vascular access for DA stenting have been described in Latin America[10,11,12]. In Brazil, this technique was reported by Arrieta et al, in a case series that included five patients with DA stenting, with ages ranging from four to nine days of life[10].

The axillary approach represents an alternative to femoral access in patients such as our patient (low-birth-weight NB) in those who are at a higher risk of femoral arterial injury. The potential advantage of the axillary approach is to access the DA with complex anatomy that does not allow an easy femoral approach. Therefore, this type of approach may be preferred for DA originated in the inferior region of the aortic arch with an acute angle[2,6,8]. Ductal stenting via the axillary route is a relatively new approach, therefore, there are only small case series showing the feasibility of this approach[X]. In our patient, the ductal stenting was successful with the complication of axillary artery spasm, which was resolved during hospitalization in intensive care. This type of complication has been described by Breatnach et al, in a study that included 20 patients of which 15% had complications in the axillary artery (2 with partial dissection, 1 pseudoaneurysm)[5]. Another additional advantage of the axillary approach is the good supply of collateral circulation of this artery, which allows protecting the femoral artery which does not offer this characteristic[18].

Although there have been advances in ductal stenting since its introduction in 1992, there are still a few failed attempts, especially when it comes to implanting it via the femoral artery in DA with abnormal origin of the aorta. In NBs, experience in these cases is even less[11,12,13,14]. Regarding the implantation of a drug-eluting stent in NBs and this case of low-birthweight, there is little experience currently available.

Up to 20 times higher levels of the drug (zotarolimus) have been reported and up to 30 times lower clearance compared with older children and adults. However, given the need to ensure that the stent remains permeable and that neointimal proliferation is reduced, the increasing use of this type of stent has been allowed even in NBs. As for the side effects or complications of these medicated stents, it has been described that the levels of immunosuppression are well tolerated in this type of patient[XI].

Recent multicenter studies comparing this percutaneous technique with surgical management have concluded that there is no difference in the primary outcome (death or unplanned reintervention to treat cyanosis); however, other markers of morbidity and pulmonary artery size favor the stented group, which supports that the ductal stent is a reasonable alternative for the surgical fistula in selected patients. The ductal stent is emerging as a preferred alternative for surgical fistula in the palliative treatment of NBs with DA-dependent pulmonary circulation because of greater stability[XII, XIII].

This case allows us to identify risks such as axillary arterial spasm, as well as the difficulties inherent to the technique of ductal stenting in DA with complex anatomy, which requires the use of additional tools such as microcatheters that allow more precise positioning of the guides and greater stability for stent advancement. The axillary arterial route is another option for the interventional approach to complex congenital heart disease. This type of approach may become more frequent as better technology with lower profile introducers becomes available and the learning curve of pediatric hemodynamics specialists increases.

Conclusion

We present the clinical case of an NB in the first week of life, with low weight, who had a stent implanted in the DA of atypical morphology (vertical), through the axillary arterial access. As an early complication of the procedure, the NB had axillary artery

spasm which resolved spontaneously. Finally, further studies are required to establish the safety of this vascular approach and its clinical implications.

Ethical Responsibilities

Human Beings and animals protection: Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

Data confidentiality: The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

Financial Disclosure

Authors state that no economic support has been associated with the

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References

1. 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.
2. Bentham JR, Zava NK, Harrison WJ, et al. Duct stenting versus modified Blalock-Taussig shunt in neonates with duct-depend pulmonary blood flow: associations with clinical outcomes in a multicenter national study. Circulation. 2018;137(6):581-8.
3. Santoro G, Capozzi G, Caianiello G, et al. Pulmonary artery growth after arterial duct stenting in completely duct-dependent pulmonary circulation: arterial duct stenting versus surgical shunt. J Am Coll Cardiol. 2009:54(23):2180-6.
4. Santoro G, Capozzi G, Caianiello G, et al. Pulmonary artery growth after arterial duct stenting in completely duct-dependent pulmonary circulation. Heart. 2016;102(6):459-64.
5. Breatnach CR, Aggarwal V, Al-Alawi K, et al. Percutaneous axillary artery approach for ductal stenting in critical right ventricular outflow tract lesions in the neonatal period. Catheter Cardiovasc. Interven. 2019;1-7. DOI: 10.1002/ccd.28302.
6. Ding L, Pocket C, Moore J, El-Said H. Long sheath use in femoral artery catheterization in infants < 15 kg is associated with a higher thrombosis rate: proposed protocol for detection and management. Catheter Cardiovasc Interv. 2016;88(7):1108-12.
7. Choundry S, Balzer D, Murpy J, Nocolas R, Shahanavaz S. Percutaneous carotid artery access in infants > 3 months of age. Catheter Cardiovasc Interv. 2016;87(4):757-61.
8. Rehman R, Marhisham MC, Alwi M. Stenting the complex patent ductus arteriosus in tetralogy of Fallot with pulmonary atresia: challenges and outcomes. Future Cardiol. 2019;14(1):55-73.
9. Riley DS, Barber MS, Kienle GS, et al. CARE guidelines for case reports: explanation and elaboration document. J Clinic Epidemiol. 2017;89:218-35.
10. Dua JS, Osborne NJ, Tometzki AjJ. Martin RP. Axillary artery approach for balloon valvuloplasty in Young infants with severe aortic valve stenosis: medium-term results. Catheter Cardiovasc Interv. 2006;68(6):929-35.
11. Lee KJ, Seto W, Benson L, Chaturvedi RR. Pharmacokinetics of sirolimus-eluting stents implanted in the neonatal arterial duct. Circ Cardiovasc Interv. 2015;8:e002233.
12. 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. Circulation. 2018;137(6):589-601.
13. 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):581-8.