

# Late transcatheter recanalization of a closed ductus arteriosus in a 2-month-old infant with tetralogy of Fallot and isolated left pulmonary artery

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## ABSTRACT

Ductal stenting is increasing as an alternative to surgical shunts in cyanotic newborns. However, most reports include newborns with patent ducts who are often on prostaglandin before the procedure. We report a successful late transcatheter recanalization of the closed ductus arteriosus in a 2-month-old infant with tetralogy of Fallot, right aortic arch, and an isolated left pulmonary artery (LPA). We achieved adequate LPA growth before complete repair. There were no procedural complications. This procedure is rare in Western countries due to universal pulse oximetry screening and increased fetal diagnoses. The procedure is technically challenging in the absence of any angiographic flow into the ductus. Evidence of a ductal ampulla increases the chances of success.

**Keywords:** Isolated pulmonary artery, patent ductus arteriosus, stent, tetralogy of Fallot

## INTRODUCTION

Ductal stenting is increasing as an alternative to early surgical shunts for ductal-dependent pulmonary blood flow. However, published reports mostly include newborns with a patent ductus arteriosus (PDA) who are often on prostaglandin before the procedure. Ductal stenting in older infants whose ducts have closed has rarely been described. In Western countries, universal pulse oximetry screening and a substantial increase of *in utero* fetal diagnoses have decreased late presenters with critical congenital heart disease. Hence, there is not much technical expertise in this scenario and no recent reports of such attempts. We report the successful transcatheter recanalization of a closed DA in a late diagnosis of tetralogy of Fallot (ToF), right aortic arch (RAA), and an isolated LPA in a Western country.

## CASE REPORT

An 8-week-old male infant weighing 6.06 kg presented with a febrile illness, cardiomegaly, and a concerning murmur. The patient's saturations were 95%–99% on room air. An echocardiogram revealed ToF with severe subvalvular and valvular pulmonary stenosis (peak gradient 96 mmHg and mean 64 mmHg). The LPA was not visualized. Cardiac computed tomography (CT) showed a severely hypoplastic isolated LPA measuring 2.5 mm (z-score, -6.22), fed entirely by small aortopulmonary collaterals. There was a RAA with mirror image branching. An ampulla at the base of the left brachiocephalic artery was suspected to be the origin of the left DA connecting to the LPA, but no blood flow was observed in the DA. The distance between the ductal ampulla and the

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LPA measured 22 mm [Figure 1]. The main and right pulmonary arteries were contiguous and normal in size. The main pulmonary artery (MPA) measured 12 mm (z-score, +0.98) and the right pulmonary artery measured 7 mm (z-score, +1.01). The pulmonary valve annulus measured 10 mm (z-score, -0.55), but was noted to be severely dysplastic by both CT scan and echocardiogram.

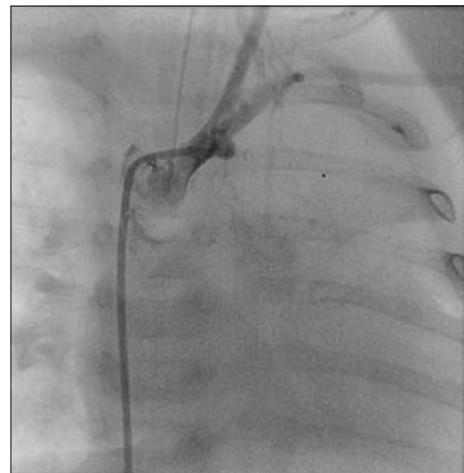
Cardiac catheterization was performed to define the native LPA and attempt ductal recanalization. A wire was advanced retrograde from the right femoral artery into the base of the ductal ampulla. No flow was noted by angiography [Figure 2 and Video 1]. Advancing a guidewire across the ductal ampulla was challenging with multiple unsuccessful attempts using various guidewires. With gentle probing, a 0.014" Floppy Guide

Wire (Boston Scientific, Marlborough, Massachusetts, the United States of America) was finally successfully advanced through the ampulla into the DA and distal LPA. Serial dilations with 2-20 and 3-20 symmetry balloons were performed at 15 atm. Repeat angiograms showed ductal recanalization and continuity from the aortic arch to the LPA with the establishment of blood flow. The LPA diameter measured 2.2 mm [Video 2a and b]. Two premounted, bare-metal coronary stents (3.5 mm × 20 mm; VeriFlex: Boston Scientific) were deployed along the entire DA. The ductus measured approximately 2.8-3.4 mm in diameter after stenting [Figure 3 and Video 3a, b].

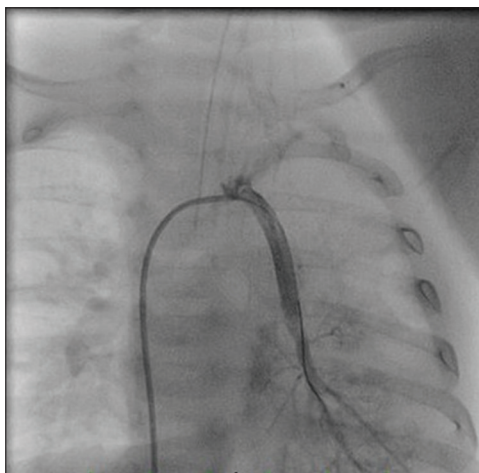
There were no perioperative complications, and the baby was discharged a week later on 40.5 mg aspirin only, which was continued till surgery. Catheterization after 3 months showed LPA growth to 5.3 mm in diameter [Figure 4 and Video 4, 5a, b]. He subsequently underwent a complete ToF repair at 5 months of



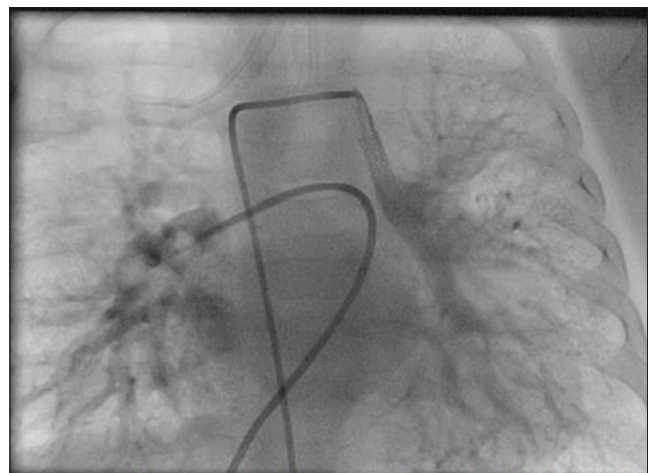
**Figure 1:** Cardiac computed tomography showing severely hypoplastic isolated left pulmonary artery fed by aortopulmonary collaterals. The distance between the ductal ampulla and the most proximal portion of the left pulmonary artery measured 22 mm in length in the coronal plane (yellow arrows). There is no flow noted in the ductus arteriosus



**Figure 2:** Cardiac catheterization shows no angiographic evidence of flow



**Figure 3:** Right aortic arch with mirror image branching. Contrast injection into the aorta shows ductal ampulla at the base of the innominate artery with no flow



**Figure 4:** Cardiac catheterization at age 6 months shows continued patency and growth of all segments of the isolated left pulmonary artery, which measured 5.3 mm in diameter

age (approximately 3.5 months after ductal stent) consisting of pulmonary valve commissurotomy, infundibular muscle bundle resection, and Dacron patch closure of the ventricular septal defect. Preoperative saturations were 98%. The duct was ligated and the stents were removed, with complete excision of the ductal tissue. MPA and LPA continuity was created by direct anastomosis posteriorly and autologous pericardial patch anteriorly. CT at 28 months showed an LPA of 12 mm. The patient is now 38 months old and thriving.

## DISCUSSION

Most reports of ductal stenting include newborns with a patent ductus who are often on prostaglandin E1 infusion before the procedure.<sup>[1,2]</sup> Recanalization of a closed PDA is reported less frequently and is a challenging procedure secondary to the difficulty of advancing a guidewire across the duct in the absence of any prograde flow across the DA (“probe patent” vs. “flow patent” DA). The high rate of *in utero* diagnosis of cyanotic heart disease in Western countries has made the need for this procedure rare, and most reports are from developing countries where the late diagnosis of critical congenital heart disease is still relatively more common. We attempted ductal recanalization to restore blood flow to an isolated and severely hypoplastic LPA after CT showed evidence of an aortic ductal ampulla. Our intervention achieved sustained flow to the LPA and adequate LPA growth with no intraprocedural complications or the need for reintervention.

Kampmann *et al.* first reported stenting a closed ductus with a 9 mm coronary stent in a 1.8 kg premature neonate also with ToF, RAA, and isolated LPA.<sup>[3]</sup> Since then, the largest report is from Kothari *et al.* who performed ductal recanalization and stenting in six patients aged 3–6 months with transposition and intact ventricular septum to achieve adequate mixing and left ventricular retraining.<sup>[4]</sup> Restenting was successful in five patients. The single failure was in an infant who did not have a definite ampulla or ductal notch on angiography. Glatz *et al.* have recently shown that adequate pulmonary artery growth may be achieved with the stenting of a PDA.<sup>[1]</sup> Our experience supports that the same may be true for the recanalization of a ductus that has already closed.

A probable surgical alternative in this patient would likely have involved staged palliation with a modified Blalock–Taussig shunt to the LPA to encourage LPA growth before full repair. Several large single and multicenter studies have reported ductal stenting to have no increased risk of mortality compared to the Blalock–Taussig shunt; rather, PDA stenting has been shown to reduce intensive care unit length of stay, greater postprocedural stability, lower postprocedural

complications, better pulmonary artery growth, and improved patient survival to destination surgical treatment.<sup>[1,5–7]</sup> Cardiac surgery post-PDA stenting has been shown to be safe and low risk. However, PDA stents may not be completely removable in all cases and may require additional pulmonary artery maneuvers during surgical repair.<sup>[8]</sup>

Our report reinforces that transcatheter recanalization and stenting of an angiographically closed DA in young infants is feasible and effective and likely supports age-appropriate pulmonary artery growth before definitive cardiac surgery. However, the procedure is technically challenging due to the difficulty of advancing a guidewire across the duct in the absence of any prograde flow across the DA (“probe patent” vs. “flow patent” DA). Chances of success may be higher in the presence of a ductal ampulla or notch. Lack of experience in such a procedure due to fewer late diagnoses of congenital heart diseases in Western countries may create a push for operative solutions or tolerance of single lung physiology. Our case report highlights the importance of attempting ductal recanalization in select cases, especially where a ductal ampulla can be seen.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

### Conflicts of interest

There are no conflicts of interest.

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