

## Stent implantation of an unusual morphology patent ductus arteriosus via Glidesheath slender

### Atipik morfolojideki patent ductus arteriozusa Glidesheath kılıf ile stent implantasyonu

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**Summary**– The procedure of stenting the patent ductus arteriosus (PDA) is a palliative procedure applied as an alternative to surgery in newborns with ductus-dependent pulmonary circulation. However, it is still a very challenging method in patients with aortic arch anomalies. We describe our experience with a newborn with right atrial isomerism and dextrocardia, complete atrioventricular septal defect, aortic outlet right ventricle with pulmonary atresia, right aortic arch, and a PDA from the left innominate artery. Because the PDA was long and tortuous, we preferred placing three short stents instead of a single long stent. The procedure applied the femoral artery approach with a Glidesheath Slender to decrease arterial injuries. PDA stenting in challenging morphologies can be performed successfully using multiple short stents and via Glidesheath Slenders.

**Özet**– Patent duktus arteriozusa (PDA) stent implantasyonu işlemi, duktus bağımlı pulmoner dolaşımı olan yenidoğanlarda cerrahiye alternatif olarak uygulanan palyatif bir işlemdir. Ancak aortik ark anomalili hastalarda halen oldukça zorlu bir yöntemdir. Bu yazıda sağ aortik arkusu olan ve PDA'sı sol innominate arterden köken alan pulmoner atrezili bir yenidoğandaki deneyimimizi paylaştık. PDA uzun ve kıvrımlı olduğu için tek bir uzun stent yerine üç kısa stent yerleştirmeyi tercih ettik. Prosedür, arter yaralanmalarını azaltmak için Glidesheath kılıf ile femoral arter yaklaşımı ile uygulandı. Atipik morfolojilerdeki PDA'larda stent implantasyonu, birden fazla kısa stent ve Glidesheath kılıf kullanılarak başarıyla gerçekleştirilebilir.

Maintaining pulmonary blood flow in infants with ductal-dependent pulmonary circulation is vital. A surgical systemic-pulmonary shunt can be a palliative solution; however, it has a significant risk of mortality and morbidity in the neonatal period.<sup>[1,2]</sup> Ductal stenting gained popularity in the early 1990s as an alternative to shunt procedures because of several advantages such as the absence of risk of phrenic or vagal paralysis, chylothorax, surgical adhesions, reduced hospital stay, and the number of reoperations.<sup>[1,3,4]</sup> Because of the significant technological advances such as smaller delivery sheaths and the pre-mounted stents designed for coronary arteries, the technique has become more preferred. However, the ductus in different morphologies can complicate the procedure, and the selection of patients, techniques, and outcomes may vary.<sup>[4,5]</sup> This brief report

described our approach to ductal stent implantation in a newborn with an infrequent PDA morphology with a right aortic arch and a PDA from the left innominate artery. To reduce vascular complications, we applied the procedure using a Glidesheath Slender, which gives the option of choosing the outer diameter one French smaller.

#### CASE REPORT

A female neonate was referred to our hospital with cyanosis on the first day of life. On physical examination, the nondysmorphic infant was cyanotic with oxygen saturations 74% on room air, 50 cm height, weighed 2.9 kg, and had a heart rate of 140 beats/min. On cardiovascular examination, the apex beat palpated at the right side of the thorax with regular

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S1 and S2 rhythm; a harsh grade 3/6 systolic ejection was also detected. A transthoracic echocardiogram revealed right atrial isomerism and dextrocardia, a complete atrioventricular septal defect (right dominant, unbalanced), an aortic outlet right ventricle with pulmonary atresia, atrioventricular valve insufficiency (moderate-severe), and a right aortic arch mirror-image branching. Moreover, confluent pulmonary arteries were retrogradely filled through a PDA originating from the left innominate artery. Prostaglandin E1 infusion was commenced, and a computed tomography angiography (CTA) scan was performed to evaluate the PDA before the invasive procedure. The CT result showed that the ductus was suitable for the femoral artery approach in terms of its location and shape (Figure 1A and B).

Catheterization was performed on day 5 after birth. PGE1 infusion was stopped 24 h before the stent implantation. General anesthesia was achieved with endotracheal intubation, and a 5F Glidesheath Slender (Terumo, Tokyo, Japan), which has an inner lumen of 5F and an outer diameter of a standard 4F sheath (which is generally 6F), was placed in the right femoral artery. A 5F sheath was placed in the right femoral vein by the percutaneous method. A dose of 200 IU heparin was administered intravenously to the patient. Antibiotic prophylaxis was performed with cefazolin for 24 h. A 5F right Judkins catheter was advanced retrogradely up to the descending aorta and PDA. An angiogram confirmed the right aortic arch mirror-image branching, and an inverted C-shaped long PDA orig-

inated from the left innominate artery and supplying confluent pulmonary arteries (Figure 1C).

**Abbreviations:**

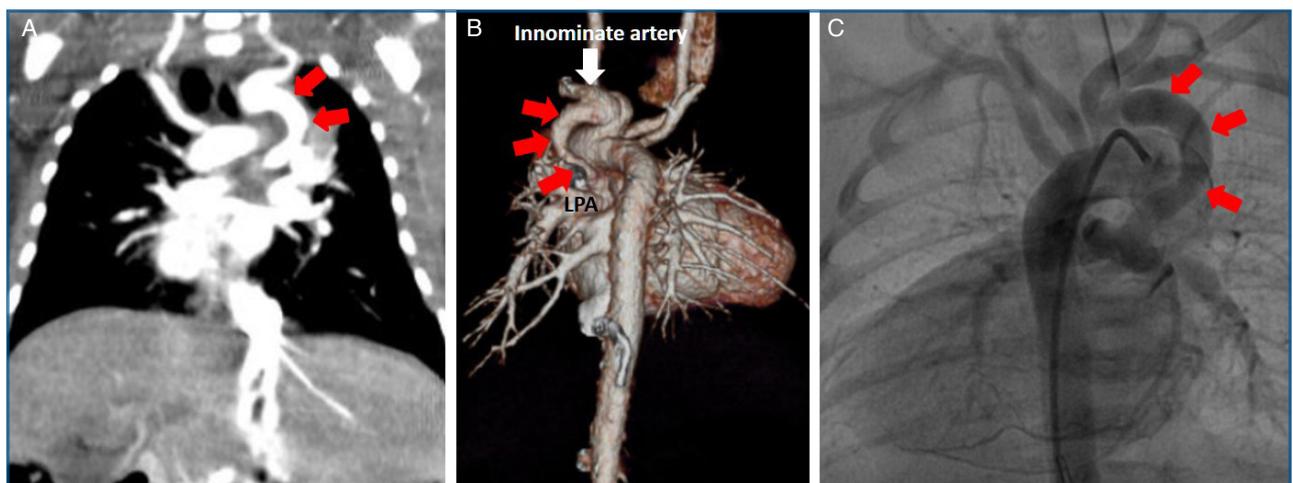
CTA Computed tomography angiography

PDA Patent ductus arteriosus

SpO<sub>2</sub> Saturated oxygen

The PDA diameter was 3 mm distally and 4.5 mm proximally; the length was estimated at 27 mm. A 5F Guiding right Judkins catheter was fed through the duct over a 0.035 in hydrophilic guidewire, and an extra support 0.014 in coronary guidewire (Iron-Man or Extra Support Abbott, Santa Clara, CA) was placed in a distal right pulmonary artery branch. The suggested stent size for patients weighing 3 kg is 4 mm.<sup>[6]</sup> Our patient was nearly 3 kg, and the ductus diameter was 3 mm at the pulmonary artery side. A 4 mm × 16 mm long coronary stent (Boston Scientific REBEL) was deployed distally in the PDA with an inflation pressure of 14 atm (Figure 2A). However, the proximal part of the ductus was larger; so, to avoid stent embolization, we preferred 4.5 mm stents for this part. A second 4.5 mm × 16 mm (Figure 2B) and a third 4.5 mm × 12 mm stents were delivered into the proximal PDA (Figure 2C). Adrenaline and milrinone infusions continued during the procedure. Immediately after stent implantation, prostaglandin infusion was stopped. In the postprocedure control injection, it was observed that the stents covered the PDA, and there was no stenosis in the distal and proximal parts (Figure 2D).

Aortic saturation increased from the previous high of 75 to 92%. Heparin infusion was commenced and

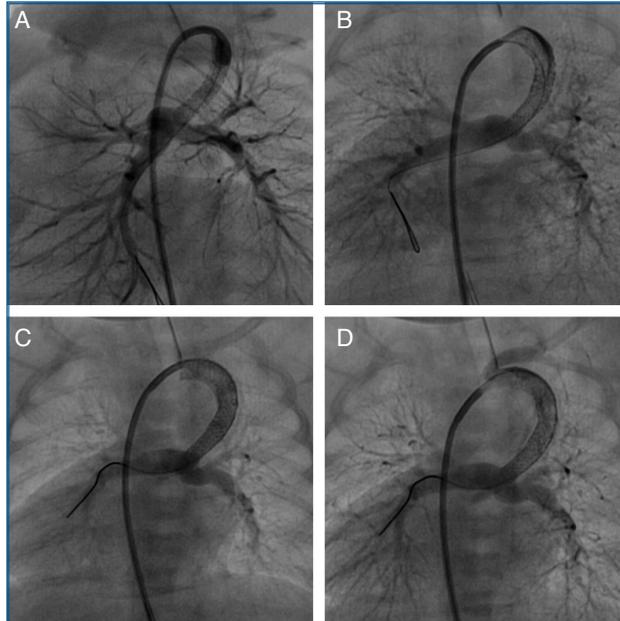


**Figure 1.** (A) CTA scan showing the tortuous PDA, (B) 3D-CTA images of the large PDA, (C) Angiogram at the origin of the left innominate artery showing large PDA (red arrows).

CTA: computed tomography angiography; PDA: patent ductus arteriosus.

administered for 24 h; aspirin and clopidogrel were started the next day and then continued with both. Echocardiography (Figure 3A) and a chest radiograph (Figure 3B) performed one day later showed the stent's unobstructed aortic and pulmonary sides. On the first day after stent implantation, we had to deal with pulmonary overflow. Saturated oxygen

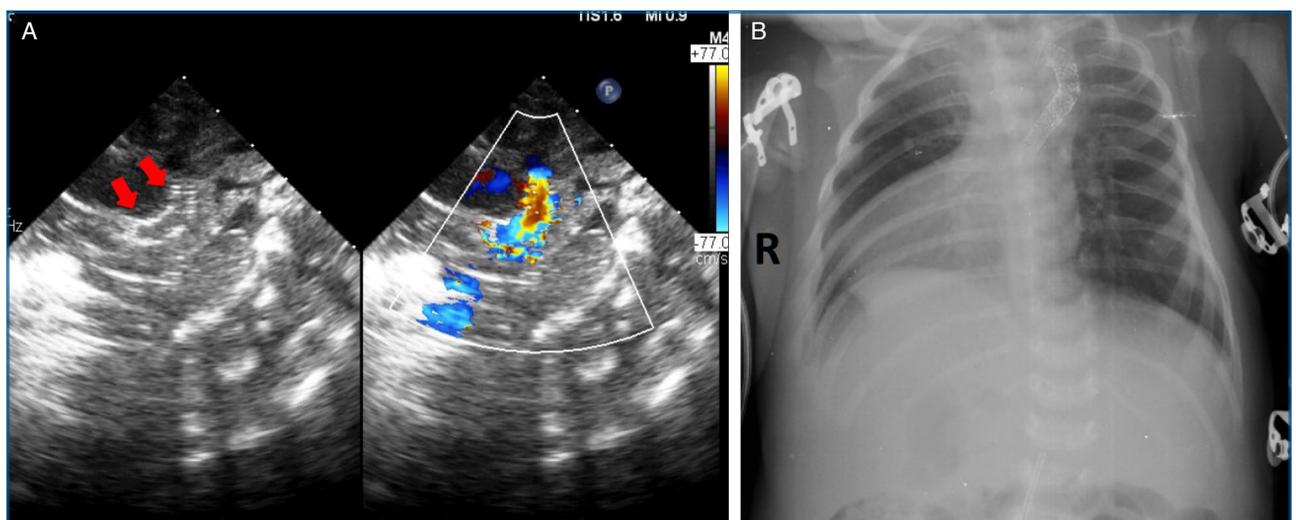
(SpO<sub>2</sub>) was nearly 100%. However, decreasing the fraction of inspired oxygen levels helped us in handling this problem. The patient was extubated after 4 days of the procedure. Unfortunately, during follow-up in the intensive care unit, first necrotizing enterocolitis and then sepsis was observed. Despite effective antibiotherapy, we could not save the baby, and she died.



**Figure 2.** (A) Angiogram in the PDA after the first, (B) after the second, and (C) after the third stent implantations. (D) Angiogram at the origin of the innominate artery showing no stenosis in the distal and proximal parts.  
CTA: computed tomography angiography; PDA: patent ductus arteriosus.

## DISCUSSION

Newborns with duct-dependent pulmonary circulation or inadequate pulmonary blood flow have traditionally been treated with a surgical shunt or have undergone early primary repair. Early primary repair is less preferred owing to the high risk of morbidity and mortality. However, systemic-pulmonary artery shunt operation may progress with significant complications, especially in premature babies. Shunt thrombosis, shunt stenosis, pulmonary or systemic arterial distortion, diaphragmatic paralysis, pleural effusion, and excessive or asymmetric pulmonary blood flow are among the most common complications.<sup>[3,7,8]</sup> Prevention of occlusion of the ductus arteriosus was considered a reasonable alternative to surgical aortopulmonary anastomosis at the end of the 1970s. In 1992 Gibbs et al.<sup>[9]</sup> described PDA stenting technique as an alternative to the systemic-pulmonary shunt. Since then, stenting of PDAs has gained increased popularity.<sup>[7]</sup>



**Figure 3.** (A) 2D and color Doppler images of the echocardiography after PDA stenting, and (B) Chest radiograph after PDA stenting.

CTA: computed tomography angiography; PDA: patent ductus arteriosus.

Usually, the PDA arises from the proximal descending aorta or the underside of the aortic arch. In patients with a right aortic arch, the PDA may originate from the innominate artery. In these patients, the PDA is generally long and tortuous; so using the closest and straightest vascular entry to the duct is critical in settling the stents. For this unusual morphology, some operators have reported ductal stenting from femoral venous approaches<sup>[4,7,10]</sup> or an arterial approach.<sup>[4,8,11]</sup> When the procedure is performed with the venous approach, the ascending aorta must be reached via the right atrium through the right ventricle through the ventricular septal defects pathway. However, especially in babies with low birth weight, hypotension and bradycardia may occur, and this situation can complicate the procedure. In this paper, we described a newborn with right atrial isomerism and dextrocardia, a complete atrioventricular septal defect (right dominant unbalanced), an aortic outlet right ventricle with pulmonary atresia, a right aortic arch, and a PDA from the left innominate artery. We implanted the ductal stent using the femoral artery approach. The arterial pathway to the PDA in this group of patients is preferable because catheter navigation is relatively easy. Nevertheless, vascular complications are the main possible concerns about the mid-term results of the procedure.<sup>[3]</sup> Hence, to prevent permanent arterial damage, we recommend the use of the smallest diameter introducer sheaths. The Glidesheath Slender is an innovative sheath with a thinner wall and hydrophilic coating. The inner diameter is compatible with 5F catheters, whereas the outer diameter is similar to that of a regular 4F sheath, which is used today in newborns for arterial access during diagnostic or interventional catheterizations. Although there are limited data presenting results of using Glidesheath Slender in small children, the initial experience with this sheath in obtaining radial access in adults is promising and shows that Glidesheath Slender does not kink more easily compared with the regular sheaths.<sup>[12]</sup> Gendera et al.<sup>[13]</sup> reported that performing percutaneous interventions in small children using the Glidesheath Slender is safe and effective. It allows for the reduction of outer sheath diameter by one French, which creates a difference in this patient group and reduces the risk of vessel complications (stenosis, occlusion). A 4F long sheath (Cook Inc, Bloomington, IN, USA) may also be preferred in patients who require PDA stenting through the femo-

ral artery.<sup>[8]</sup> However, especially in PDAs originating from the innominate artery, as it will be difficult to advance the long sheath to that region, it can be performed through the 5F guiding catheter. Another advantage of using a guiding catheter is that it is easier to manipulate than a long sheath when more than one stent is placed.

It is essential to cover the entire length of the ductus in the aortic stump to prevent ductal stenosis and avoid potential injury from the stent edges. So, multiple stents may be used to establish a curve concordant to the ductus course.<sup>[1,2,7]</sup> In short and straight PDA cases, stent length is relatively easy to determine by angiography; however, it can be challenging in a tortuous ductus where the length is not precisely predictable. In these cases, choosing a moderately longer stent than the ductal size and placing multiple short stents instead of long stents is recommended.<sup>[1]</sup> Choosing a single long stent can cause technical difficulties. Roggen et al.<sup>[4]</sup> reported three unsuccessful ductal stent cases with ductus arteriosus from the innominate/subclavian artery. Kinking of the long stent was the reason for failure in the first case. The second failure was because of the long stent, which could not be advanced into the PDA. In the last one, PDA straightening occurred during balloon inflation and caused the stent to shift in the ductus because of its long length. Three stents were placed in our patient, starting at the pulmonary side by the telescoping technique; thus, the ductus line without stenosis could be established.

In long and tortuous PDAs, such as the one in our brief report, it will be safer to place multiple short stents instead of a single stent. This process can be an alternative to aortopulmonary shunt surgery. Besides, interventions through the Glidesheath Slender in small patients are safe and feasible even when using an arterial approach.

**Informed consent:** Informed consent was obtained from the patient for the publication of the case report and the accompanying images.

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**Keywords:** Ductus arteriosus stenting; innominate artery; right aortic arc; Glidesheath

**Anahtar Kelimeler:** Duktus arteriozusa stent; innominate arter; sağ aortik arkus; Glidesheath