CASE REPORT

# Rare combination and transcatheter treatment during single session in an infant: Patent ductus arteriosus and major aortopulmonary collateral artery concordance

Patent duktus arteriyozus ve majör aortopulmoner arter birlikteliği:
Nadir bir birliktelik ve süt çocuğu döneminde
aynı seansta kateter yoluyla tedavisi

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Summary— Major aortopulmonary collateral arteries are abnormal vascular structures that may be seen in cyanotic diseases that progress with reduced pulmonary flow. They occur rather rarely in the absence of cyanotic congenital heart disease. Presently described is the case of an infant who underwent patent ductus arteriosus (PDA) and major aortopulmonary collateral artery occlusion in a single session, without presence of cyanotic congenital heart disease. To the best of our knowledge, this is the first case of congenital aortopulmonary collateral artery to be reported in a symptomatic infant with PDA.

Özet- Majör aortopulmoner kollateral arterler, pulmoner akım azlığıyla giden siyanozlu hastalıklarda görülebilen anormal damarsal yapılardır. Siyanozlu doğumsal kalp hastalığı olmadığı durumlarda oldukça seyrek görülürler. Bu yazıda, siyanozlu doğumsal kalp hastalığı olmayan, aynı seansta patent duktus arteriyozus (PDA) ve majör aortopulmoner kollateral arter tıkaması uygulanan bir süt çocuğu sunuldu. Bildiğimiz kadarıyla bu olgu, PDA'sı olan semptomlu bir süt çocuğunda doğumsal aortopulmoner kollateral arter birlikteliği olan ilk olgudur.

Major aortopulmonary collateral arteries (MAPCAs) are collateral vascular formations that may occur in concomitance with insufficient pulmonary circulation. [1] MAPCAs are unexpected when sufficient blood flow is provided to the pulmonary vascular bed.

Presently described is the case of an infant who underwent patent ductus arteriosus (PDA) and MAP-CA occlusion in the same session, when no cyanotic congenital heart disease was present.

## **CASE REPORT**

The male patient was born in the 37<sup>th</sup> week, and was 42 days old. Monitored in the neonatal intensive care

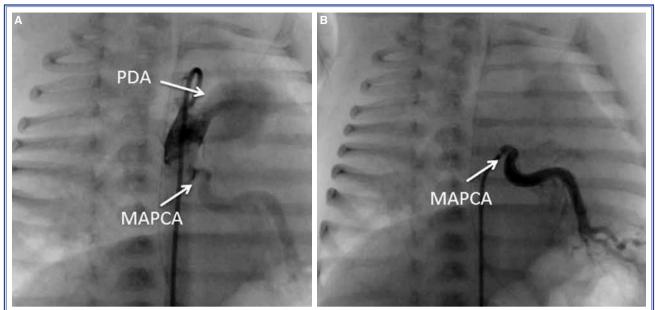
unit, the patient was unable to achieve independence from mechanical ventilator and failed to gain weight. O, saturation was 98%, and

#### Abbreviations:

MAPCA Major aortopulmonary collateral arteries PDA Patent ductus arteriosus

a 3/6 systolic-diastolic murmur was detected in the upper left part of the sternum upon auscultation. On transthoracic echocardiography, wide, left-to-right shunted PDA, left atrial dilatation, and left ventricular dilatation were identified. The patient had significant pulmonary hypertension, and systolic heart functions were normal. Suprasternal exposure revealed a MAP-CA originating from the descending aorta. The intubated patient was transferred to the catheter hall. After contrast media injection into the descending aorta, a

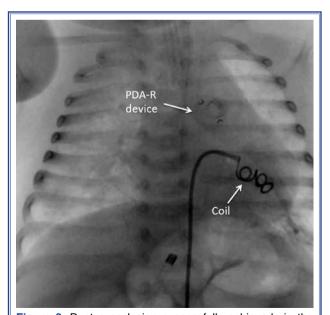




**Figure 1.** Following contrast media injection into the descending aorta, **(A)** a wide PDA and MAPCA were observed, as was **(B)** a MAPCA extending from the descending aorta to the lower left lung lobe.

PDA measuring 4.9 mm at its narrowest, 11 mm at the ampulla, and 8 mm in length was detected, as was a MAPCA extending from the descending aorta to the lower left pulmonary lobe (Figure 1).

Ductus occlusion was successfully performed using a PDA-R occluder 7 mm in diameter (Produkte für



**Figure 2.** Ductus occlusion successfully achieved via the venous route using an occluder device 7 mm in diameter. The distal part of the collateral artery was accessed via the arterial route, and the coil was released in the middle segment of the collateral artery.

die Medizin AG, Cologne, Germany) and a 4-F JR4 catheter. A microcatheter was then advanced through a 4-F Cobra catheter via the arterial route, and the distal part of the collateral artery was accessed using a coronary guidewire. A 5×4-mm coil was released in the middle segment of the collateral artery. As passage was present during control injection, a second, 3×3-mm coil was released into the collateral artery (Figure 2). When contrast injection was repeated, the collateral artery was completely occluded, and no significant residue was present. The patient was admitted to the intensive care unit, and was extubated after 2 days. On control transthoracic echocardiography it was observed that dilatation in the left cardiac chambers had decreased, pulmonary hypertension had disappeared, and congestion in the lungs had thoroughly improved.

#### DISCUSSION

MAPCAs are embryonic ventral splanchnic artery residues that may occur in conjunction with cyanotic diseases that accompany tetralogy of Fallot, ventricular septal defect-pulmonary atresia, and pulmonary stenosis. These abnormal vascular structures may heterogeneously increase pulmonary blood flow in different segments of the lungs, result in volume burden on the left ventricle, lead to recurrent lung infection, and cause heart failure. MAPCAs may also

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develop in isolation secondary to hypoxia, inflammation, or trauma, particularly in premature infants without concomitant congenital heart disease. [3] Generally speaking, they frequently present as small, asymptomatic arteries in patients with bronchopulmonary dysplasia. [2]

A MAPCA accompanying PDA was presently observed in the absence of cyanotic congenital heart disease progressing with reduced pulmonary blood flow. In spite of prolonged intubation, the patient had a wide PDA, increased pulmonary blood flow, and pulmonary hypertension. We believe that prolonged intubation was not an influential factor, and that the development of MAPCA was coincidental. Recently, interventional treatment options have been used, as surgical ligation of these vascular structures is challenging. Coils and vascular plugs are frequently used in transcatheter MAPCA embolization. In the present case, MAPCA occlusion with 2 coils was successfully achieved via the arterial route.

In conclusion, MAPCA is a collateral vascular formation that is generally expected in cases of cyanotic heart disease with reduced pulmonary blood flow. However, it may also accompany certain heart pathologies with rich pulmonary blood supply, as in the present case. Transcatheter treatment performed

in the same session in patients with heart pathologies that require multiple interventions may provide satisfactory results.

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### **REFERENCES**

- Sharma S, Kothari SS, Krishnakumar R, Saxena A, Sharma R, Taneja K, et al. Systemic-to-pulmonary artery collateral vessels and surgical shunts in patients with cyanotic congenital heart disease: perioperative treatment by transcatheter embolization. AJR Am J Roentgenol 1995;164:1505–10. Crossref
- Patra S, Srinivas SK, Agrawal N, Jayaranganath M. Isolated major aortopulmonary collateral artery in an infant presenting with recurrent lower respiratory tract infection. BMJ Case Rep 2013. Crossref
- Padhi SS, Bakshi KD, Shastri RK. Multiple coil closure of isolated aortopulmonary collateral. Ann Pediatr Cardiol 2010;3:65–7. Crossref

Keywords: Infant; major aortopulmonary collateral artery; patent ductus arteriosus; transcatheter treatment.

Anahtar sözcükler: Bebek; majör aortopulmoner kollateral arter; patent duktus arteriyozus; transkateter tedavi.