Clinical Case Report

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Patent ductus arteriosus coexisting with a left brachiocephalic artery originating from the descending aorta

A case report

Jiabing Huang, MD, PhD, Xiaofan Peng, MD, PhD, Xiangqian Shen, MD, Xinqun Hu, MD, PhD, Zhenfei Fang, MD, PhD*

Abstract

Rational: Patent ductus arteriosus (PDA) and a coexisting left brachiocephalic artery originating from the descending aorta is an extremely rare anomaly of unknown etiology.

Patient concerns: Herein we report a 3-year-old female who was found to have this condition during intervention process to close PDA.

Diagnosis: The patient was diagnosed with PDA coexisting with left brachiocephalic artery through angiography.

Intervention: Intervention involved transcatheter closure of the pulmonary side of PDA with coils.

Outcomes: At 6-months follow up, the patient was well, with no symptoms and normal flow through the left carotid artery.

Lessons: PDA coexisting with left brachiocephalic artery originating from the descending aorta is a very rare anomaly. When this variety of PDA is closed, it is important to avoid affecting the blood flow in the left brachiocephalic trunk. For this reason, closure on the side of the pulmonary artery may be the best solution

Abbreviations: PDA = patent ductus arteriosus, RAA = right aortic arch.

Keywords: left brachiocephalic artery, patent ductus arteriosus, transcatheter closure

1. Introduction

Patent ductus arteriosus (PDA) is among the most common congenital heart defects. In the fetus, the ductus arteriosus is connected via the descending aorta to the main pulmonary trunk; normally it closes soon after birth. Persistence of ductal patency results in left-to-right shunting of blood from the aorta to the pulmonary artery. This leads to pulmonary hypertension and can eventually cause right-to-left shunting (Eisenmenger's syndrome) as well as other complications, such as congestive heart failure, endarteritis, and aneurysm of the ductus arteriosus.

Branching of the aortic arch varies. Over 50% of individuals have 3 aortic branches (brachiocephalic trunk, left common carotid, and left subclavian artery), whereas some have 4 (brachiocephalic trunk, left common carotid, and left and right subclavian arteries). In rare cases, the left and right brachiocephalic trunks arise from the aortic arch or the left common carotid artery originates from the brachiocephalic trunk.[1] Aortic arch branching patterns fall into 3 primary types: double aortic arch, right aortic arch with mirror-image branching and no concomitant cardiac abnormalities, and right aortic arch with abnormal branching.[2]

It is very rare to detect a left brachiocephalic trunk originated from the descending aorta. The patient reported herein was a 3-year-old female who presented with a PDA coexisting with a left brachiocephalic trunk originated from the descending aorta.

2. Case report

This study was approved by Ethical Review Committee of the Second Xiangya Hospital of Central South University (Changsha, Hunan, China) and written informed consent was obtained from the patient's parents. Their 3-year-old child was admitted to the department of cardiology because of her cardiac anomaly, which had been diagnosed a week earlier. Echocardiography suggested that she had a PDA, and the diameter of her pulmonary artery was slightly larger than usual. After routine examinations before intervention, percutaneous closure of the PDA was performed under general anesthesia. Two 5-Fr sheaths were embedded in the right femoral vein and artery after they were punctured. A 5-Fr pigtail catheter was passed through the ascending aorta and

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Department of Cardiovascular medicine, the Second Xiangya Hospital of Central South University, Changsha, China.

** Correspondence: Zhenfei Fang, Department of Cardiovascular medicine, the Second Xiangya Hospital of Central South University, No. 139, middle Ren-min road, Changsha, Hunan 410011, China (e-mail: fangzhenfei@csu.edu.cn).*

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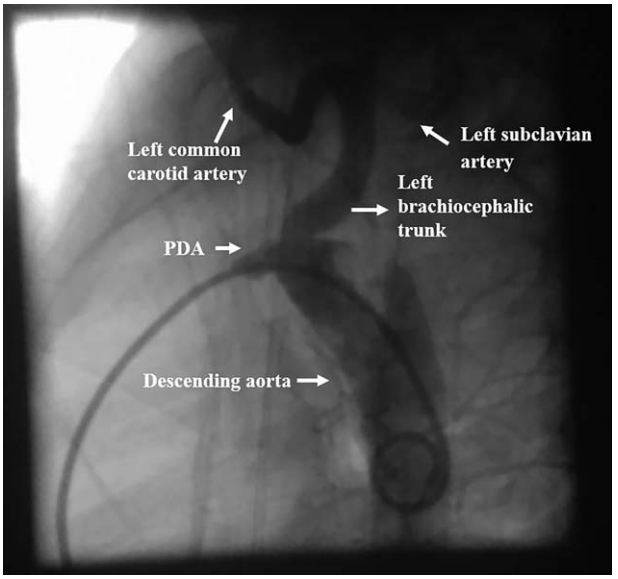

Figure 1. Lateral projection angiogram of the descending aorta shows a PDA and left brachiocephalic trunk branching into the left common carotid artery and left subclavian artery. The origins of the left brachiocephalic trunk and PDA are the same. PDA = patent ductus arteriosus.

descending aorta from different access points in order to perform angiography, followed by right heart catheter examination. The left brachiocephalic trunk (branching into the left common carotid artery and left subclavian artery) arising from the descending aorta, in addition the origin of the left brachiocephalic trunk, were found to be on the aortic side of the PDA (Figs. 1 and 2). A continuous pressure curve from the ascending aorta to the descending aorta revealed that there was no pressure gradient between the 2 parts of the aorta. We utilized a coaxial guide

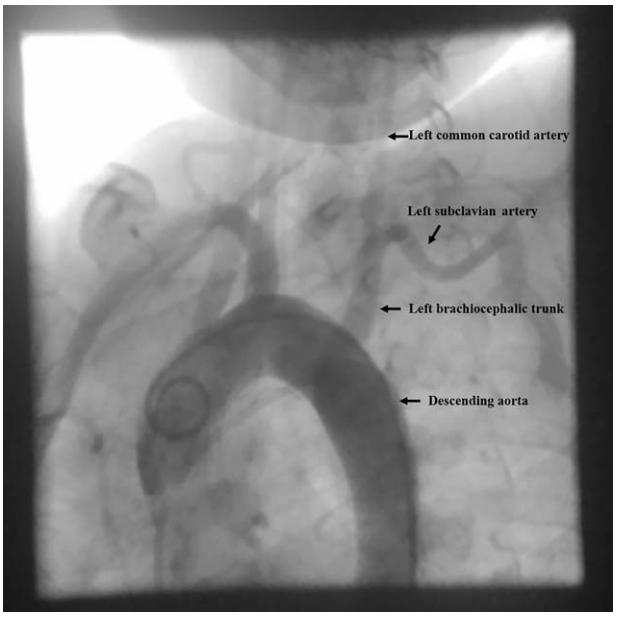

Figure 2. Posteroanterior projection angiogram of the ascending aorta shows the left brachiocephalic trunk stemming from the descending aorta.

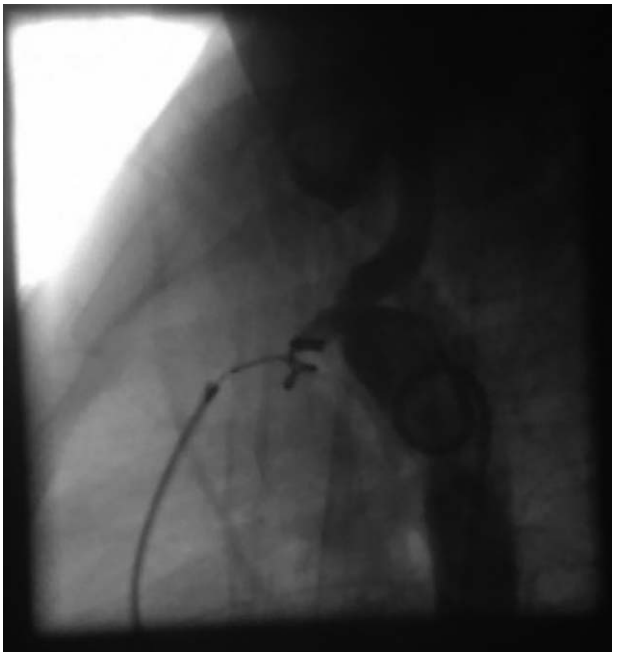

Figure 3. Lateral projection angiogram of the descending aorta after coils were deployed confirms complete occlusion of the PDA, but blood flow in the left brachiocephalic trunk has not been impaired. PDA = patent ductus arteriosus.

system with an outer 5-Fr sheath and inner 5-Fr 100-cm end-hole coil delivery catheter (5-Fr multipurpose catheter, Cordis, Johnson, Miami, Florida) across the PDA with access from the right femoral vein, after which a coil delivery system (IWMCE-5-PDA3, detachable coil, Cook Medical, Bloomington, Indiana) was inserted into the multipurpose catheter and advanced into the PDA. Once the coil delivery system was in position, the coil was deployed. Angiography performed through the pigtail catheter then confirmed complete occlusion of the ductus with no effect on flow in the left brachiocephalic artery (Fig. 3). At 6-month follow-up, the patient was well, with no symptoms and normal flow through the left carotid artery.

3. Discussion

During cardiovascular growth, the proximal and distal portion of 6 pair of embryonic aortic as proximal branch pulmonary arteries and ductus arteriosus respectively. The latter links the left pulmonary artery with the left dorsal aorta.[3] The duct is essential to pulmonary growth during the fetal period. Normally the distal right sixth aortic arch loses its connection to the dorsal aorta and degenerates. This transformation is usually complete by 8 weeks of fetal life. Persistence of ductal patency after birth, connecting the aorta and pulmonary artery, results in a shunt connecting the systemic circulation with the pulmonary circulation. Left-to-right shunting through the ductus leads to pulmonary over circulation and a volume overload in the left heart. In addition, in patients with large shunts, this causes decreased lung compliance, which can give rise to increased work of breathing. With long-standing left-to-right shunting, increased pulmonary artery pressure and flow bring about progressive morphologic changes in the pulmonary vasculature, including endometrial hyperplasia, tube wall thickening, and luminal stenosis until finally pulmonary vascular resistance exceeds

systemic vascular resistance. At this point the left-to-right shunt changes to a right-to-left shunt, or Eisenmenger syndrome, which is a terminal stage of congenital heart disease. Our patient had left ventricular enlargement, which called for closure of the PDA.

Most people have a normal aortic arch branching into the brachiocephalic trunk, left common carotid, and left subclavian arteries from right to left. Nevertheless, some variants of this arrangement have been found, especially in conjunction with cardiac anomalies. A right aortic arch (RAA) is among the most common variants.[4] RAA is frequently associated with monoallelic deletion of chromosome 22q11 regardless of the presence of associated cardiac anomalies.[5] McElhinney et al[2] has published a systemic review of patterns of RAA morphology and mirror-image branching of the brachiocephalic vessels. In this article, we can see that RAA with mirror-image branching often coexists with PDA or with tetralogy of Fallot, pulmonary atresia, or truncus arteriosus.

However, PDA without other cardiac anomalies combined with an aortic variant is rare. Manyama et al[6] has reported a case manifesting coexistence of a PDA and left brachiocephalic artery. In addition, there is our case, reported here, present a case of PDA coexisting with a left brachiocephalic artery originating from descending aorta adjacent to the paten ductus arteriosus. This appears to be the first such case to appear in the literature. Radiological examination is the main method to diagnosis the anomaly. Posteroanterior projection of aortic angiogram can clearly exhibit the PDA and aortic arch, which easily reveals not only the origin of anomalous artery but also the morphology, trail, branches and region of blood supplement.

Our patient presented with a left common carotid artery originating from the proximal part of the left third aortic arch, whereas the left subclavian artery derived from the left seventh intersegmental artery. Why the left brachiocephalic trunk derived from the descending aorta is uncertain. We conjecture that the distal part of the left sixth embryonic aortic arch has 2 branches, one of which evolves into the right brachiocephalic trunk and the other into the ductus and eventually the left brachiocephalic trunk. The latter may be associated with transforming growth factor beta-SMAD2 signaling in aortic development during the fetal period.[7]

Routine PDA closure technique utilizes coils placed at the aortic side of the PDA from femoral artery access. However, in this case, we adopted a rare procedure using femoral vein access to block the pulmonary side of the PDA, thus avoiding thrombus caused by the coils, which could have affected blood flow through the left brachiocephalic trunk.

4. Conclusion

In summary, PDA coexisting with a left brachiocephalic trunk derived from the descending aorta originating from the arterial side of the PDA is a rare anomaly. When this variety of PDA is closed, it is important to avoid affecting the blood flow in the left brachiocephalic trunk. For this reason, closure on the side of the pulmonary artery may be the best solution.

Author contributions

Conceptualization: Zhenfei Fang.
Data curation: Xiaofan Peng.
Methodology: Xiangqian Shen.
Supervision: Xinqun Hu.
Writing – original draft: Jiabing Huang.
Writing – review & editing: Jiabing Huang.

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