

Aortic Coarctation 28 Days after an Arterial Switch Operation in a Neonate

Fumiaki Shikata, MD
Toru Okamura, MD
Takashi Higaki, MD
Masahiro Okura, MD
Ai Kojima, MD
Shunji Uchita, MD
Hironori Izutani, MD

Aortic coarctation rarely occurs after an arterial switch operation for D-transposition of the great arteries with intact ventricular septum. We report the case of a neonate patient in whom aortic coarctation developed 28 days after an uncomplicated arterial switch operation. Preoperatively, the aorta was noted to have an irregular shape, but there was no pressure gradient across the lesion. The patient underwent successful reoperation to correct the coarctation. We hope that our report raises awareness of a rare early complication after arterial switch operation with intact ventricular septum, and the need to carefully monitor the aortic isthmus in patients who have aortic irregularities, even in the absence of a pressure gradient. (Tex Heart Inst J 2016;43(4):354-6)

Key words: Abnormalities, multiple/surgery; aortic coarctation/etiology/surgery; cardiac surgical procedures/adverse effects; infant, newborn; postoperative complications/diagnosis; reoperation; transposition of great vessels/complications/surgery; treatment outcome; ventricular septum/physiology

From: Departments of Cardiovascular Surgery (Drs. Izutani, Kojima, Okamura, Okura, Shikata, and Uchita) and Pediatric Cardiology (Dr. Higaki), Ehime University Hospital, Ehime 791-0295, Japan

Address for reprints: Fumiaki Shikata, MD, Department of Cardiovascular Surgery, Ehime University, Shitsukawa, Toon, Ehime 791-0295, Japan

E-mail: shika@blue.ocn.ne.jp

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Case Report

Right ventricular (RV) outflow obstruction and aortic insufficiency after an arterial switch operation (ASO) are not unusual, and patients in whom these complications develop need to undergo reoperation. On the other hand, aortic coarctation develops infrequently after an ASO for transposition of the great arteries (TGA) with intact ventricular septum.¹⁻³ Even when no obstructive lesions are noted in the aorta before the ASO, coarctation can occur at any time, even later than a year postoperatively. Aortic coarctation during the early postoperative stage has been reported very infrequently.^{1,2,4} We report the case of a neonate in whom aortic coarctation developed 4 weeks after an uncomplicated ASO for D-transposition of the great arteries (D-TGA) in the presence of an intact ventricular septum.

In April 2013, a female infant was delivered at 39½ weeks' gestation (birth weight, 3.125 kg), and she was transferred to our institution because of significant cyanosis. After admission, echocardiograms revealed TGA, an intact ventricular septum, and a patent foramen ovale. The aorta was seen to be anterior to the pulmonary artery. After the diagnosis was confirmed, a continuous infusion of prostaglandin E₁ was started to maintain patency of the ductus arteriosus (DA). A catheter examination revealed a coronary pattern classified as ILCx-2R and no aortic pressure gradient, although the isthmus was noted to have a somewhat irregular shape (Fig. 1A). Echocardiograms showed no abnormal flow through the aortic isthmus. There was no subaortic stenosis, and the diameters of the aortic and pulmonary valve annuli at birth were 8.21 mm (Z-score, 2.05) and 8.71 mm (Z-score, 0.9), respectively.

Thirteen days after birth, the patient underwent an ASO. Cardiopulmonary bypass (CPB) was established, and the DA was ligated and divided at the position most distant from the descending aorta. The prostaglandin infusion was discontinued. The aorta was cross-clamped, and a solution of glucose, insulin, and potassium was infused. The coronary arteries were transferred to each neo-aortic sinus. The aorta was unclamped, and the pulmonary trunk was reconstructed with use of the Lecompte procedure and a fixed autologous pericardial patch. The duration of CPB was 207 min, and the aortic cross-clamp time was 135 min. The patient was weaned uneventfully from CPB and was transferred to the intensive care unit (ICU) in relatively stable condition. At that time, a transthoracic echocardiogram showed no aortic coarctation, and arterial pressure lines inserted in the patient's upper and lower limbs revealed no pressure gradient. Echocardiograms showed preserved left ventricular function and no pulmonary or aortic stenotic lesions, so the patient was extubated and moved to the neonatal care unit for observation.

Twenty-eight days postoperatively, a stenotic aortic isthmus was identified on a contrast-enhanced computed tomogram (Fig. 1B), and the difference in blood pressures between the patient's upper and lower limbs was 30 mmHg. Therefore, urgent coarctation repair was performed. The patient's chest was opened through a left 3rd intercostal thoracotomy. The aorta was dissected between the proximal portion of the left subclavian artery and the descending aorta, and the distal and proximal sites of the aorta were clamped. Of note, in the stenotic portion, the divided site of the DA was of adequate distance from the stenotic portion of the aorta (Fig. 1C)—indicating that iatrogenic stenosis had not caused coarctation. Aortic tissue from the coarctation lesion and ductal tissue were carefully removed, and the aorta was reconstructed by means of an extended end-to-end anastomosis. After the aorta was unclamped, resolution of the coarctation was confirmed through arterial lines that were inserted into the patient's upper and lower limbs. The patient's chest was closed, and she was transferred to the ICU. She recovered uneventfully after the 2nd operation and was discharged from the hospital 3 weeks later. At the 3-year follow-up evaluation, she was in good condition and had no residual coarctation.

Discussion

Early results of the ASO have improved because of better surgical techniques and perioperative care; however, late complications such as neo-aortic insufficiency and RV outflow obstruction—which might necessitate reoperation—can occur in some patients.³ Aortic coarctation after an ASO is less likely, even though the preoperative coexistence of aortic coarctation and TGA or double-outlet RV is relatively prevalent.⁵ Three sug-

gested causes of aortic coarctation after an ASO are compression of the aorta by the surgical clips during closure of the patent DA; kinking in the aorta from compression associated with the Lecompte procedure during reconstruction of the pulmonary arteries; and contraction of the ductal tissue that extends to the proximal aorta.^{1,2,4,6,7} Presumably, contraction of our patient's ductal tissues could have caused aortic coarctation to develop earlier after ASO than in the other reported cases. Moreover, this patient's maintenance on prostaglandin E₁ before the ASO might have prevented contraction of the ductal tissue and the early coarctation.

When the aortic arch is abnormal, as seen on the preoperative angiogram in our patient (Fig. 1A), the postoperative development of aortic coarctation should be anticipated. In such cases, a trial of stopping prostaglandin in the ICU, with continuous use of echocardiography and close monitoring of 4-extremity blood pressures, would be the best way to evaluate whether aortic coarctation might develop early after ASO after the patent DA has been closed. This is particularly important when an irregularity of the aortic isthmus is identified, despite no pressure gradient across the aortic lesion on preoperative examination. Egan and associates⁴ and Muster and colleagues⁷ suggested that an irregular isthmus before surgical repair is a possible risk factor for coarctation, which can develop at any time after ASO. This hypothesis has been supported by autopsy findings in which patterns of the aortic arch predicted the development of coarctation regardless of whether ductal tissues had closed spontaneously.⁸ In addition, Egan and associates⁴ suggested that infusing prostaglandin can decrease the pressure gradient through an irregular aortic isthmus. Consequently, when a structural irregularity of an aortic isthmus is preoperatively identified despite no pressure gradient across the lesion, surgeons

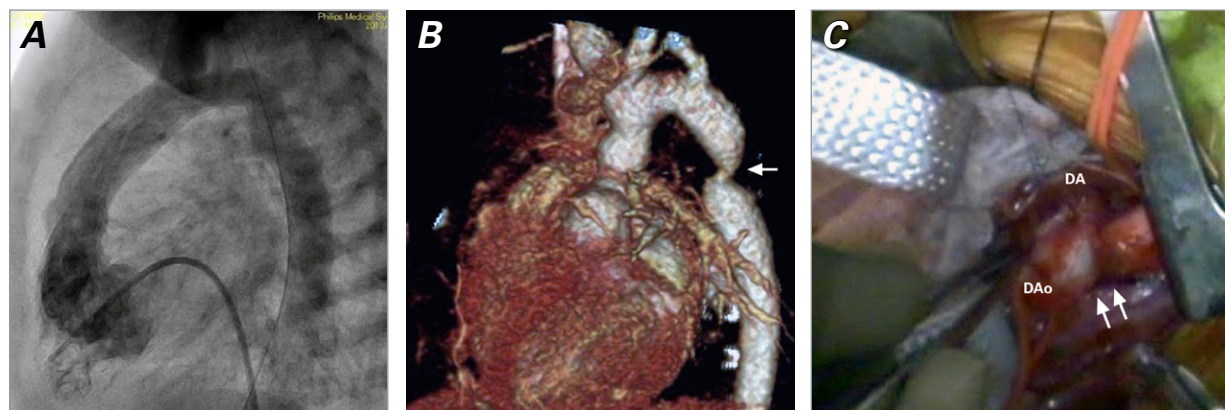


Fig. 1 **A)** Preoperative angiogram shows an irregular lesion in the aortic isthmus, without a pressure gradient. **B)** Contrast-enhanced computed tomogram after the arterial switch operation shows a severely narrowed, newly developed aortic coarctation. **C)** Photograph during reoperation shows aortic coarctation in the juxtaductal portion. Arrows indicate the stenotic segment of the aorta.

DA = ductus arteriosus; DAo = descending aorta

need to continuously monitor patients preoperatively, because concomitant coarctation repair might be indicated during the initial surgery.

We agree that the aortic isthmus should be carefully monitored after an ASO for D-TGA with intact ventricular septum, even when an aortic stenotic lesion has not been observed in the presence of preoperative prostaglandin administration and an irregularity of the aortic isthmus. We hope that our report raises awareness of this possible early complication in patients similar to ours.

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