

## CASE REPORTS

## Rescue Coronary Artery Bypass Grafting in a Neonate After Arterial Switch Operation: A Temporary or a Lifetime Solution?

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**ABSTRACT** We describe a case where prompt myocardial rescue revascularization at the time of arterial switch operation (ASO) for D-transposition of the great arteries (D-TGA) was fundamental for overcoming the acute intraoperative myocardial injury and for restoring prompt left ventricular function. Five years following the ASO the patient was asymptomatic but signs of myocardial ischemia were found at SPECT MI, potentially leading to ischemic complications. Adult patients who required coronary revascularization in the neonatal period need to be followed for life to prevent and treat any possible cause of further myocardial ischemia during childhood and adulthood. doi: 10.1111/jocs.12565 (*J Card Surg* 2015;30:611–613)

Arterial switch operation (ASO) with coronary translocation is a well-established surgical technique for patients with D-transposition of the great arteries (D-TGA).<sup>1</sup> Additional surgical techniques, which require manipulation and reimplantation of coronary arteries, can be adopted in patients with D-TGA, especially in cases with abnormal origin and courses of coronary vessels or in cases of iatrogenic coronary lesions.<sup>2–3</sup> Coronary artery bypass grafting (CABG) is considered a good surgical option for patients who had an iatrogenic coronary lesion after coronary translocation that did not resolve after conventional surgical maneuvers.<sup>4–7</sup>

**Conflict of interest:** none

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We present a case of a neonate with D-TGA, ventricular septal defect, and abnormal coronary artery pattern who underwent successful rescue CABG for acute ischemia at the time of ASO. Early and long-term clinical results are detailed.

### Case presentation

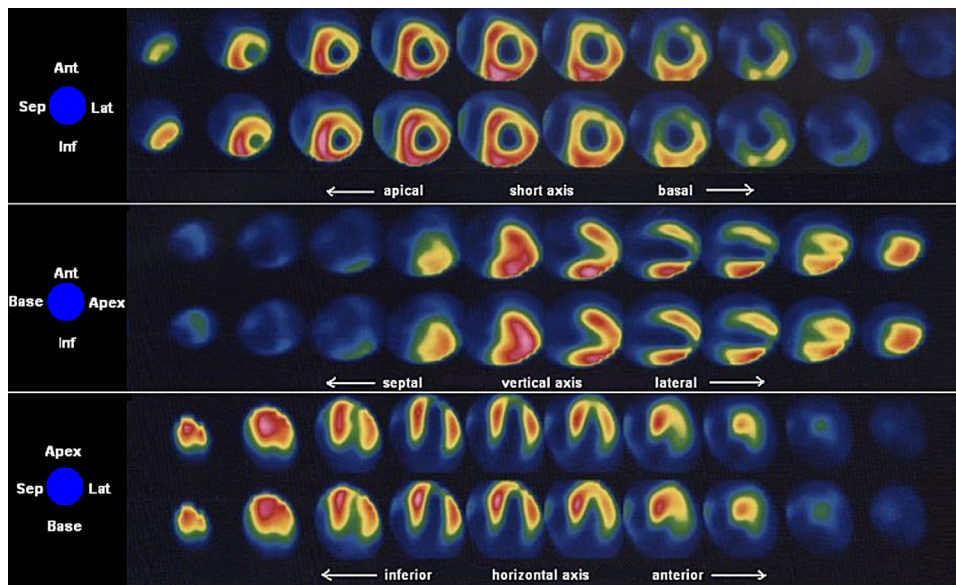
Institutional review board permission was obtained to report this case. An 11-day-old neonate with D-TGA and ventricular septal defect (VSD) was scheduled for an ASO. A preoperative 2D-echo examination was performed showing a normal coronary artery pattern. Intraoperatively inspection of the anatomical structures revealed the presence of coronary anomalies characterized by a single ostium coronary origin from the right coronary sinus and an abnormal intramural course of the left main coronary artery (LMCA). After the unroofing of the LMCA origin, we were able to detach two separate coronary buttons which were subsequently reimplanted on the neo-aorta. On weaning from the cardiopulmonary bypass (CPB), an area of myocardial discoloration corresponding to the LMCA territory was noted followed by EKG changes (ST elevation in V2-V5). CPB was reestablished and the left internal mammary artery (LIMA) was harvested and employed, as a pedicle graft, to perform a CABG on the LMCA with an immediate restoration of myocardial perfusion. We used interrupted 8.0 polypropylene sutures for the LIMA anastomosis. The patient was then weaned successfully from CPB and had an uneventful postoperative course.

The 2D echo examination at discharge (on postoperative day 22) revealed good overall ventricular ejection fraction and absence of mitral and aortic regurgitation. A rest and stress SPECT myocardial perfusion imaging (MPI) showed no areas of myocardial ischemia. The patient was then discharge home on oral aspirin (5 mg/kg/day).

Five years following ASO the patient is asymptomatic, leading a normal life with a regular exercise tolerance and a good LV function at 2D-echo examination. A rest and stress EKG revealed no ischemic changes. A follow-up stress SPECT myocardial image (MI) shows reduced perfusion on the basal, medium, and anterior segments of the LV (Fig. 1). Coronary angiography showed a complete occlusion of the previous CABG and a severe stenosis of the LMCA ostium and collateral circulation from the right coronary artery (Fig. 2).

### DISCUSSION

Coronary complications and myocardial revascularization techniques have been extensively described after ASO for D-TGA.<sup>4–9</sup> Surgical procedures in neonates, which require reimplantation and manipulation of coronary arteries, may be associated with potential serious complications especially in case of abnormalities of origin, position, or distribution of these arteries.<sup>1,9</sup>



**Figure 1.** Stress SPECT MI scan image showing a lack of distribution of the radiopharmaceutical medication of contrast on the anterior and lateral regions of the left ventricular myocardium.

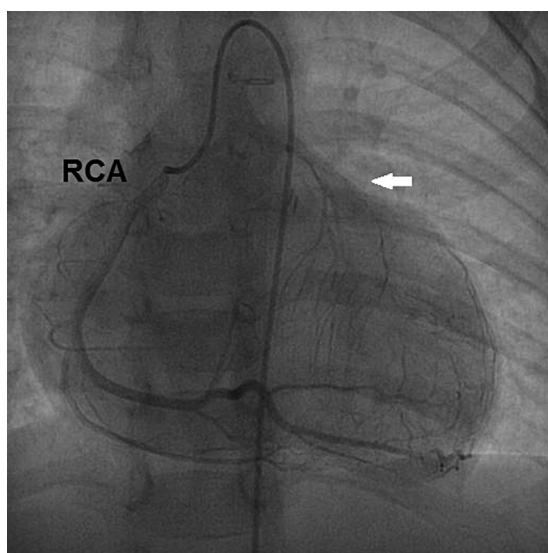
During the last two decades, CABG has been described as a successful rescue option in neonates with D-TGA who had coronary artery ischemia at the time of ASO and has favorably impacted the clinical course of those patients with coronary insufficiency.<sup>4-6</sup> In particular, the utilization of the left internal thoracic artery for CABG in children seems to have the higher patency rate as well as demonstrated anastomotic and linear growth potential.<sup>7</sup> Nonetheless there is still concern about its patency in patients younger than three years of age.<sup>4,8</sup> Moreover, it is well known that children rarely report symptoms and noninvasive

methods have not been predictive of coronary abnormalities even when a severe coronary lesion is present thus making the follow-up and the diagnosis of myocardial ischemia or myocardial perfusion impairment challenging.<sup>4</sup>

We described a case with intraoperative diagnosis of coronary anomalies, where a prompt myocardial rescue revascularization at the time of ASO was fundamental for overcoming the acute intraoperative myocardial injury and for restoring a proper left ventricular function. The intraoperative finding of anomalous coronary origin or course is rare since more than 98% of coronary patterns are currently diagnoses by preoperative 2D-echo examination. As a consequence a routine preoperative imaging study including a CT or angiography to delineate the coronary anatomy is not routinely performed in our institution.

At the time of surgery, the creation of two coronary buttons, after the unroofing of the LMCA from the single ostium coronary system, was particularly challenging, and this was the reason why we decided to perform a CABG following the finding of LMCA territory ischemia at the time of CPB discontinuation, rather than reinspecting our unroofing procedure to find any potential site of stenosis on the LMCA. We believe that CABG failed due to a progressive stenosis at the anastomotic site with consequent thrombosis of the internal thoracic artery.

Myocardial revascularization in the neonatal period is a reliable rescue solution for improving immediate patient survival. However, due to the unclear long-term graft patency, these patients needed to be followed closely for life to evaluate the occurrence of symptoms and to prevent and treat any possible cause of further myocardial ischemia during childhood and adulthood. Our patient was still asymptomatic five years after ASO, with good left ventricular function on 2D-echocardiography; however, signs of myocardial



**Figure 2.** Angiographic image of the aorta (anterior-posterior view) with selective right coronary injection showing a well developed intercoronary collateral circulation from the right coronary artery. RCA: right coronary artery. White arrow: indicating the left coronary artery.

ischemia were noted on SPECT MI, followed by angiographic confirmation of CABG occlusion. Given the relatively high incidence (8-10%) of occluded native coronaries after ASO<sup>1,2,4,9</sup> and due to the fact that symptoms, EKG, and 2D-echo seem unreliable to detect a "silently occluded" graft, a yearly SPECT MI is highly recommended, especially in patients with coronary artery anomalies or requiring other intraoperative coronary maneuvers at the time of ASO.

The right timing for coronary reoperation in case of silent ischemia after neonatal CABG still needs to be clarified, as well as the most appropriate surgical strategy (including surgical coronary ostium plasty versus a redo-CABG), as well as the choice of the most appropriate conduits for a re-vascularization procedure (i.e., the use of saphenous veins), especially in infants and adolescents.

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## Lymphangioma Presenting as Cardiac Tamponade in a Child

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**ABSTRACT** Lymphangiomas are hamartomatous malformations, which are commonly encountered in children. Intrapericardial lymphangioma is exceptionally rare. We present a case of an intrapericardial lymphangioma in a child presenting with cardiac tamponade. doi: 10.1111/jocs.12572 (*J Card Surg* 2015;30:613–616)

In children, increased accumulation of the fluid due to trauma, inflammation, and hypothyroidism are well-known causes of cardiac tamponade.<sup>1</sup> We report a case of cardiac tamponade in a 30-month-old male due to an intrapericardial cystic multiloculated mass, which on histopathology showed features of a lymphangioma.

## CASE REPORT

A 30-month-old male presented with complaints of rapidly progressive dyspnea for three days. The physical examination revealed facial puffiness, dilated neck veins, tachycardia, and muffled heart sounds. Chest radiograph revealed an enlarged cardiac silhouette (Fig. 1A). A two-dimensional echocardiography revealed compression of the heart by an intrapericardial multiloculated cystic mass. A computed tomographic (CT) scan showed a 12 × 9 cm well-defined hypodense cystic collection with multiple thin septa (Fig. 1B), displacing the pulmonary trunk posteriorly. An emergency surgery was planned.

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