

Bilateral internal thoracic artery grafting in children under 5 years of age with Kawasaki disease: a case series

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Background

Although persistent coronary artery aneurysm of Kawasaki disease (KD) is rare, some patients develop a life-threatening myocardial infarction. In paediatric coronary artery bypass surgery (PCABS), the internal thoracic artery (ITA) graft is a reliable graft with favourable coronary outcomes. However, few studies have reported the outcomes and technical considerations of PCABS using bilateral ITAs in small children who have multivessel disease.

Case summary

We present the cases of three children under 5 years of age who underwent PCABS utilizing bilateral ITAs. All three patients had known bilateral giant coronary aneurysms associated with KD. Paediatric coronary artery bypass surgery was indicated after confirming multiple coronary lesions with myocardial ischaemia. One child underwent emergency PCABS because of circulatory collapse. The arterial anastomoses were performed under cardioplegic arrest. The left ITA was anastomosed to the left anterior descending artery (LAD) or the circumflex artery. The right ITA was anastomosed to the right coronary artery or the LAD. Post-operative coronary angiography revealed patent bilateral ITA grafts with an excellent run-off in all patients, and none of them have suffered any subsequent coronary event.

Discussion

Bilateral ITA grafting is a feasible procedure with favourable coronary outcomes for treating small children with multivessel disease, even in the setting of circulatory collapse.

Keywords

Kawasaki disease • Small children • Paediatric coronary artery bypass surgery • Bilateral internal thoracic artery grafting • Case report

Learning points

- Bilateral internal thoracic artery grafting is a feasible procedure with favourable coronary outcomes for treating small children with multivessel disease-associated Kawasaki disease.
- The techniques required for paediatric coronary artery bypass surgery in small children are also applicable in infants and neonates who experience coronary complications during congenital cardiac operations.

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Introduction

The reported incidence of Kawasaki disease (KD) differs between countries. In European studies, the incidence is 5–10 per 100 000 children under 5 years of age.¹ Total annual hospitalization rates have shown a decreasing trend in the incidence of KD over the past 10 years in the USA (18.0 per 100 000 children under 5 years of age in 2012).² Conversely, its incidence has increased in Japan (309.0 per 100 000 children under 5 years of age in 2016).³ Giant coronary aneurysms associated with KD are less likely to regress to normalized arteries, resulting in the possible development of a severe stenotic lesion just proximal or distal to the aneurysm during a long-term period. Although persistent coronary artery aneurysms are uncommon, with an incidence of ~5%, some patients develop a life-threatening myocardial infarction due to the occlusion of the perianeurysmal stenotic lesion or thrombosis.⁴ Paediatric coronary artery bypass surgery (PCABS) utilizing the internal thoracic artery (ITA) is an essential intervention in KD patients who have significant coronary stenotic lesions because the ITA has a growth capability and offers long-term graft patency.⁵ However, few studies have reported the outcomes or technical considerations of bilateral ITA grafting in small children with multivessel disease.

Timeline

Patient	Age	Examination	Events, Findings
1	2 months	Transthoracic echocardiography (TTE)	Development of Kawasaki disease Bilateral coronary artery aneurysms
	4 years and 8 months	Coronary angiography (CAG)	Post-aneurysmal stenosis in the right coronary artery (RCA) and post-aneurysmal occlusion in the left anterior descending artery (LAD) and the circumflex artery
	4 years and 10 months	Single-photon emission computed tomography (SPECT) Computed tomography angiography (CTA)	Left ventricular ejection fraction (LVEF) 38%, infarction, and ischaemia in the anteroseptal wall Aneurysms in the RCA, the left main trunk, and the proximal LAD

Continued

Continued

Patient	Age	Examination	Events, Findings
	6 years	CAG SPECT	Suddenly circulatory collapse 1 day before a scheduled paediatric coronary artery bypass surgery (PCABS) Emergency PCABS [left internal thoracic artery (ITA)-LAD, right ITA-RCA] in 2009 Patent bilateral ITA grafts with an excellent run-off LVEF 46%, perfusion defect in the anterior and the posterolateral wall, no residual ischaemia
	14 years	CTA Cardiac magnetic resonance imaging (CMR) TTE	Widely patent bilateral ITA grafts LVEF 41%, endocardial scar in the anterolateral wall, transmural mottled scar in the septum, and the inferior wall LVEF 54%, no mitral regurgitation No coronary events
2	2 years and 1 month	TTE	Development of Kawasaki disease, bilateral coronary artery aneurysms
	3 years and 1 month	CAG SPECT CTA	Post-aneurysmal stenosis in the LAD and the RCA LVEF 64%, infarction and ischaemia in the anteroseptal and apical wall Aneurysms in the RCA and proximal LAD
	3 years and 8 months		PCABS (left ITA-LAD, right ITA-RCA) in 2011

Continued

Continued

Patient	Age	Examination	Events, Findings
	7 years	CTA	Patent bilateral ITA grafts
	8 years	CAG SPECT	Widely patent bilateral ITA grafts with excellent run-off LVEF 74%, a localized perfusion defect in the anterior wall, no residual ischaemia
	10 years	TTE	LVEF 67%, no mitral regurgitation
3	11 years		No coronary events
	8 months	TTE	Development of Kawasaki disease Bilateral coronary artery aneurysms
	2 years and 8 months	CAG	Occlusion in the proximal LAD, collaterals from the RCA to the LAD
	2 years and 10 months	CAG	Development of acute myocardial infarction in the lateral wall Post-aneurysmal occlusion in the left main coronary artery
	2 years and 11 months	SPECT	LVEF 58%, infarction and ischemia in the inferolateral wall PCABS (left ITA-circumflex artery, right ITA-LAD) in 2018
	3 years	CAG SPECT CMR	Patent bilateral ITA grafts with excellent run-off LVEF 73%, no perfusion defect, no residual ischaemia LVEF 55%, endocardial scar in the anterolateral wall
	4 years		No coronary events

Case presentation

From 2000 to 2018, we managed three patients under 5 years of age who underwent PCABS by utilizing bilateral ITAs. Pre-operative transthoracic echocardiography showed no mitral regurgitation in all patients. The patients were under cardiopulmonary bypass with cardioplegic arrest. All anastomoses were created in an end-to-side fashion with continuous 8-0 polypropylene sutures. Surgical glasses with an $\sim 4\times$ power magnifying lens were used. A longitudinal incision of 4–5 mm was created at the anastomotic site of the coronary artery, and the end of the ITA was longitudinally cut back 6–8 mm.

Patient 1

In 2009, a 4 year and 10 months old boy (body surface area of 0.70 m², bodyweight of 15.5 kg), had coronary ischaemia resulting from known bilateral calcified giant coronary aneurysms. He had an occlusive left main coronary artery and 75% stenosed right coronary artery (RCA) (Figure 1A). Myocardial perfusion single-photon-emission computed tomography (SPECT) revealed infarction and ischaemia in the anteroseptal region with a left ventricular ejection fraction (LVEF) of 38%. One day before a scheduled PCABS, just after suffering chest discomfort, he suddenly developed circulatory collapse with bradycardia requiring cardiopulmonary resuscitation and was rushed to the operation room. After the quick establishment of cardiopulmonary bypass, bilateral ITAs were harvested in the semi-skeletonized fashion. The left ITA was anastomosed to the left anterior descending artery (LAD) with a 1.25-mm shunt tube. The right ITA was anastomosed to the distal RCA with a 1.5 mm shunt tube because the left circumflex artery was small with sparse opacification by contrast medium in pre-operative coronary angiography (CAG) and a possible cause of bradycardia was developing RCA occlusion (Figure 1B). Cardiopulmonary bypass was discontinued, and he was transferred to the intensive care unit with the sternum open. Delayed sternal closure was performed 5 days post-operatively. He was extubated at 24 days and was discharged from our centre 131 days post-operatively. Coronary angiography conducted 1 year later showed patent grafts with excellent run-off and nearly occluded coronary aneurysms (Figure 1C). Computed tomography angiography (CTA) at 6 years showed widely patent grafts. Myocardial perfusion SPECT revealed a perfusion defect in the anterior and the posterolateral wall, and no residual ischaemia with an LVEF of 46%. Ten years post-operatively, echocardiography revealed an improved LVEF of 54%. Currently, he leads a normal life at school with moderate-intensity physical activities.

Patient 2

We were following-up a 3 year and 8 months old boy (body surface area of 0.56 m², bodyweight of 12.8 kg), for giant coronary aneurysms with critical stenotic lesions in the LAD and 90% stenotic lesions in the RCA (Figure 2A). In 2011, he underwent PCABS because a myocardial perfusion SPECT revealed infarction and ischaemia in the anteroseptal and apical regions with an LVEF of 64%. Bilateral ITAs were harvested in the semi-skeletonized fashion. The left ITA was anastomosed to the LAD with a 1.25 mm shunt tube. The right ITA

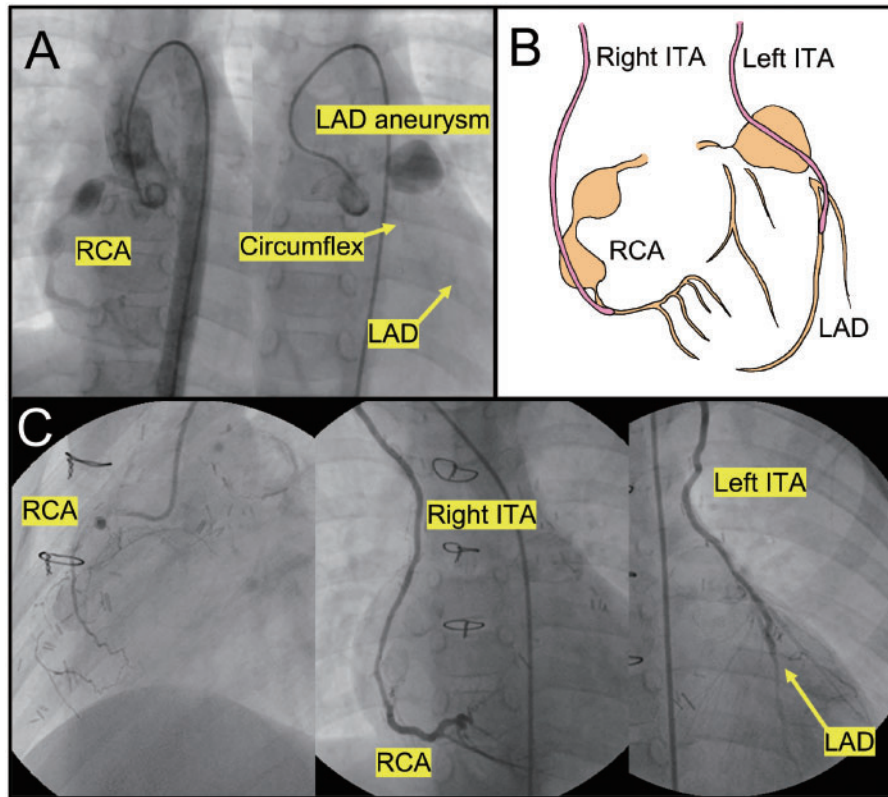


Figure 1 (A) Pre-operative coronary angiography. Post-aneurysmal occlusion in the left anterior descending artery and the circumflex artery, and post-aneurysmal stenosis in the right coronary artery. Sparse opacification by the contrast medium in the left anterior descending artery and the circumflex artery. (B) Operative illustration. (C) Coronary angiography conducted 1 year post-operatively. Patent internal thoracic artery grafts with excellent run-off and nearly occluded coronary aneurysms.

was anastomosed to the distal RCA with a 1.25 mm shunt tube (Figure 2B). He was extubated on the day of the operation and was discharged from our centre 17 days post-operatively. A CAG at 4 years showed widely patent grafts with excellent run-off and occluded coronary aneurysms (Figure 2C). Myocardial perfusion SPECT revealed a localized perfusion defect in the anterior wall and no residual ischaemia with an LVEF of 74%. Eight years post-operatively, he leads a normal life at school.

Patient 3

In 2018, a 2 year and 11 months old girl (body surface area of 0.58 m², bodyweight of 13.5 kg), developed an acute myocardial infarction in the lateral wall during the follow-up of known bilateral coronary aneurysms. Coronary angiography showed a nearly occluded aneurysm in the left main coronary artery with occluded lesions in the LAD and the circumflex artery. A giant coronary aneurysm in the proximal RCA had no stenotic lesions (Figure 3A). Myocardial perfusion SPECT revealed infarction and ischaemia in the inferolateral wall with an LVEF of 58%. One month later, she underwent PCABS. Bilateral ITAs were harvested in the skeletonized fashion. The left ITA was anastomosed to the LAD with a 1.5 mm shunt tube. The right ITA was anastomosed to the obtuse marginal branch with a 1.0 mm shunt tube (Figure 3B). She was extubated 1 day post-

operatively, and discharged from our centre 18 days later. One month post-operatively, CAG showed patent grafts with excellent run-off (Figure 3C). At 7 months, myocardial perfusion SPECT revealed significantly improved coronary perfusion in the inferolateral region and no residual ischaemia with an LVEF of 73%. One year post-operatively, the CTA showed widely patent grafts, and currently, she leads a normal life.

Discussion

In small-bodied children, PCABS utilizing the ITA is technically demanding.⁶ Harvesting of the ITA is relatively easy with the skeletonized or the semi-skeletonized fashion, however, the wall of the ITA is thin, and the lumen is ~1 mm in diameter or less. Furthermore, the distal end of the ITA may shrink because the ITA of small children has an excellent elasticity. Coronary artery walls are also thin and the lumen diameter ranges from 1.25 mm to 1.5 mm in the mid-portion of the LAD and the RCA. Because a meticulous anastomosis is required, the use of surgical glasses with over 4× magnifying lens and cardioplegic arrest are useful in making the anastomotic lumen as wide as possible with a small suturing bite. A coronary shunt tube helps to secure the anastomotic lumen. It also

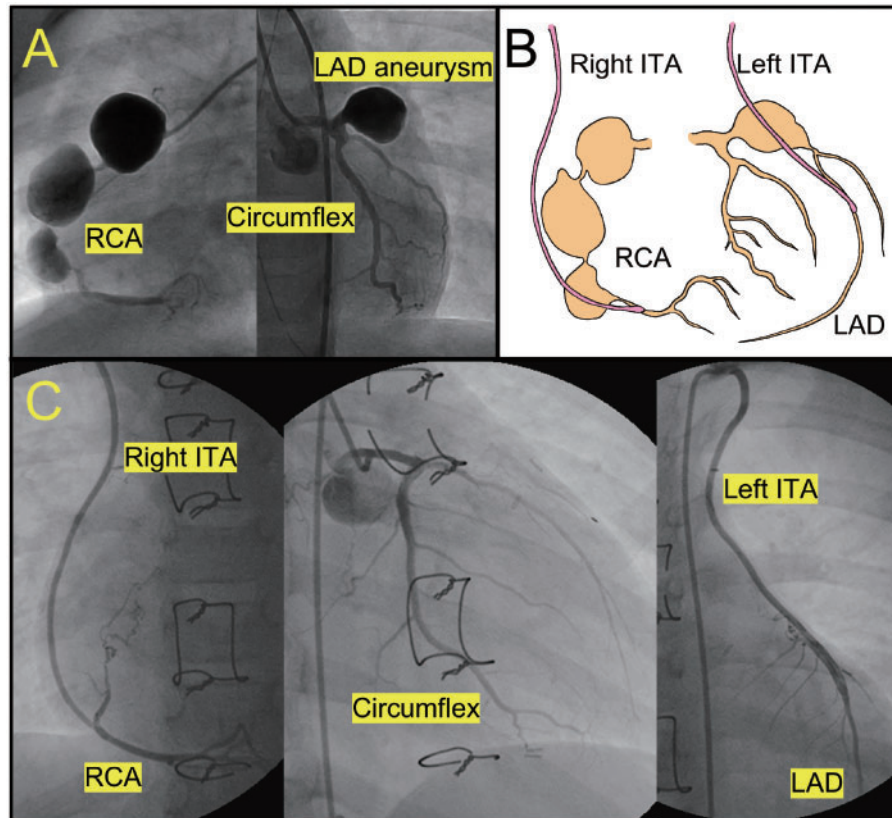


Figure 2 (A) Pre-operative coronary angiography. Post-aneurysmal stenosis in the right coronary artery and critical post-aneurysmal stenosis with delayed opacification by the contrast medium in the left anterior descending artery. (B) Operative illustration. (C) Coronary angiography conducted 4 years post-operatively. Widely patent internal thoracic artery grafts with excellent run-off and occluded coronary aneurysms.

helps to prevent suturing the posterior and opposite wall of the artery at the anastomotic site.

In 1990, Kitamura *et al.*⁶ first reported successful bilateral ITA grafting in eight children ranging from 3 to 13 years old. Bilateral ITA grafting has since gradually become a standard procedure in multivessel PCABS. We previously reported that the 20-year patency rate of the ITA was 87%,⁵ and the 15-year cardiac event-free rate was 84.1%⁷ for KD patients who underwent coronary artery bypass surgery. However, most of the patients in these studies were aged 5 years or more. Cooley *et al.*⁸ reported an ITA graft in a 16-month-old child, which remained patent 10 years post-operatively. Mavroudis *et al.*⁹ reported favourable mid-term survival in three children under 5 years of age who underwent PCABS with an ITA graft. Kitamura *et al.*⁶ reported a 13-month good outcome in a 3-year-old child who underwent PCABS utilizing bilateral ITA grafts. In the present report, bilateral ITA grafting contributed to improved coronary blood flow and an increase in the LVEF. The patients were able to lead a normal life without any coronary events. These cases suggest that bilateral ITA grafting to two of three coronary territories is effective for preserving cardiac function through to adulthood, even in small children with multivessel disease resulting in circulatory collapse.

The techniques required for PCABS are also applicable in infants and neonates who experience coronary complications during congenital cardiac operations.¹⁰ Mavroudis *et al.*¹¹ reported angiographic measurements of the coronary arteries of 34 children, including neonates. The mean sizes of the LAD, RCA, and left ITA were 1.21 mm, 1.33 mm, and 1.18 mm, respectively, in neonates. In infants, they were 1.25 mm, 1.39 mm, and 1.21 mm, respectively. They also reported a patent left ITA to LAD graft 52 days post-operatively in a 4-month-old female infant who had critical left coronary ostial stenosis.¹¹ D'Saouza *et al.*¹² reported a 7-week-old male neonate with left main coronary artery atresia and mitral regurgitation who underwent a left internal thoracic artery (LITA) to LAD bypass and mitral valve repair. Coronary angiography 1 year post-operatively showed a patent left ITA graft with excellent run-off. Nair *et al.*¹³ reported a successful bilateral ITA to LAD and RCA bypass in a 23-week-old infant who underwent an arterial switch operation. These reports confirm that even in neonates and infants, the ITA and coronary arteries are 1 mm or larger, and ITA to coronary artery bypass with proper PCABS technique is a feasible and effectively life-saving procedure.

In conclusion, PCABS utilizing bilateral ITA grafts is an effective procedure with favourable coronary outcomes in small children with multivessel disease.

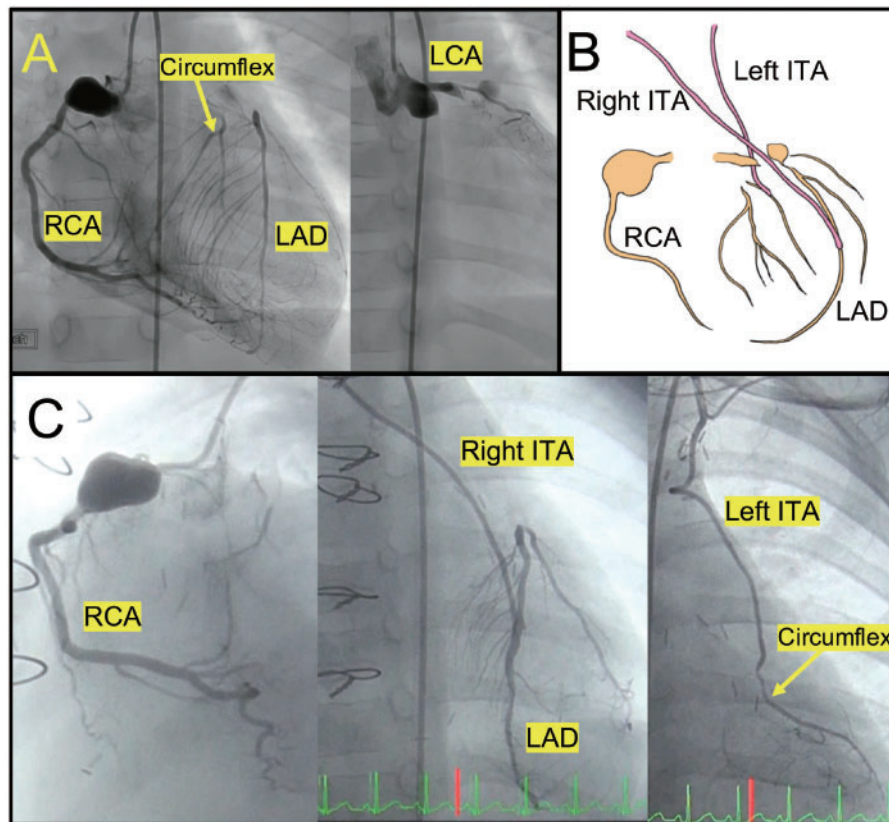
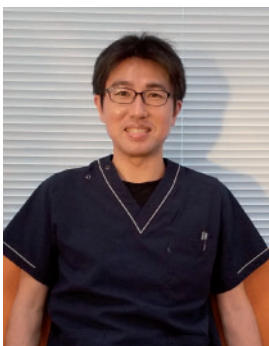


Figure 3 (A) Pre-operative coronary angiography. Post-aneurysmal occlusion in the left main coronary artery. Opacification of the left anterior descending artery and circumflex artery by the contrast medium from collaterals of the right coronary artery. (B) Operative illustration. (C) Coronary angiography conducted 1 month post-operatively. Patent internal thoracic artery grafts with excellent run-off.

Lead author biography



Yusuke Shimahara is a cardiovascular surgeon at National Cerebral and Cardiovascular Center, Osaka, Japan. He earned his PhD degree from Tohoku University in Sendai, Japan. He has a lot of expertise in cardiac surgery including off-pump coronary artery bypass surgery, valvular surgery, and transcatheter aortic valve replacement. His research interests include all arterial aortic no-touch off-pump coronary artery bypass surgery, hybrid off-pump coronary

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Supplementary material

Supplementary material is available at *European Heart Journal - Case Reports* online.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as [Supplementary data](#).

Consent: The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patients' next of kin in line with COPE guidelines.

Conflict of interest: none declared.

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