Cardiology in the Young

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Brief Report

Cite this article: Asada D, Tomita H, Fujii T. (2019) Successful everolimus-eluting stent implantation into the left main trunk stenosis in the anomalous coronary artery after neo-aortic valve replacement in a 6-year-old boy. *Cardiology in the Young* **29**: 448–450. doi: 10.1017/S1047951118002512

Received: 25 May 2018
Revised: 31 October 2018
Accepted: 6 December 2018
First published online: 4 February 2019

Key words:
Drug-eluting stent; Norwood operation; neo-aortic valve replacement; anomalous coronary artery

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Successful everolimus-eluting stent implantation into the left main trunk stenosis in the anomalous coronary artery after neo-aortic valve replacement in a 6-year-old boy

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Abstract

CHDs occasionally have coronary complications; however, stent implantation is technically difficult in small children. We reported a successful drug-eluting stent implantation into the congenital anomalous coronary artery in a 6-year-old boy. This treatment is useful for rescuing coronary stenosis, and dual antiplatelet therapy is important to prevent stent restenosis.

Despite several case reports describing successful deployment with acceptable short-term experiences in small children,[1] coronary stent implantation is generally not considered a standard treatment option owing to its technical difficulties. It becomes more difficult in the anomalous coronary artery.

Here, we reported a successful stent deployment into the left main trunk stenosis in a 6-year-old boy with coronary artery angiectopia after neo-aortic valve replacement.

Case report

The patient was a 6-year-old boy weighing 20 kg. He was diagnosed with false Taussig–Bing anomaly, right ventricular hypoplasia, and aortic arch interruption and had Shaher type 9 coronary arterial anatomy (Fig 1a).[2] He underwent bilateral pulmonary artery banding, Norwood operation, and bidirectional cavo-pulmonary shunt in stages at his previous hospital. At the age of 1-year, he was referred to our hospital for total cavo-pulmonary connection; however, transthoracic echocardiography revealed severe neo-aortic regurgitation. He underwent total cavo-pulmonary connection at the age of 2 years, but the neo-aortic regurgitation gradually worsened and advanced atrioventricular block emerged. Neo-aortic valve replacement using 21-mm-diameter On-X valve (CryoLife Inc., Kennesaw, GA) and pacemaker implantation were performed at the age of 6 years. After this surgery, transthoracic echocardiography indicated low ejection fraction of 25% by Simpson method and hypokinesis of the anterior and lateral left ventricular wall. Electrocardiography showed ST-T alteration and Q wave in the precordial leads (Fig 2a), indicating ischemic change. We performed coronary angiography to investigate the ischemia. The standard view was unable to clearly detect the coronary orifice, but the coronary anatomy was well detected in the caudal view (Fig 1b). Coronary angiography revealed 90% stenosis of the left main trunk compressed by the mechanical valve; this stenosis was considered the cause of cardiac dysfunction. We decided to implant a drug-eluting stent to rescue the coronary blood flow. Informed consent was obtained from the parents.

Stent implantation was performed under general anaesthesia. A right femoral vascular access was established, and 100 U/kg of intra-arterial heparin was administered. A 6-F Judkins left guiding catheter (Heartrail II; Terumo Corp, Tokyo) was advanced into the orifice of the left coronary artery, and a 0.014 in. guidewire (SION blue; Asahi Intecc Co, Nagoya) was inserted into the left main trunk. Intravascular ultrasonography (OptiCross™; Boston Scientific Corp., Marlborough, MA, United States of America) indicated a minimal lumen diameter of 0.4 mm and reference vessel diameter of 2.5 mm. Angiography revealed a stenosis length of 14 mm. We chose a 2.5–18 mm drug-eluting stent (XIENCE ALPINE; Abbott Vascular, Chicago, IL, United States of America) and deployed it at 10 atm. Post-dilation was performed using a 2.5–10 mm balloon catheter (Raiden 3; KANEKA MEDIX, Osaka) at 24 atm. Final angiography showed adequate dilation and good stent position (Fig 2b). No ischemic events occurred during the procedure.

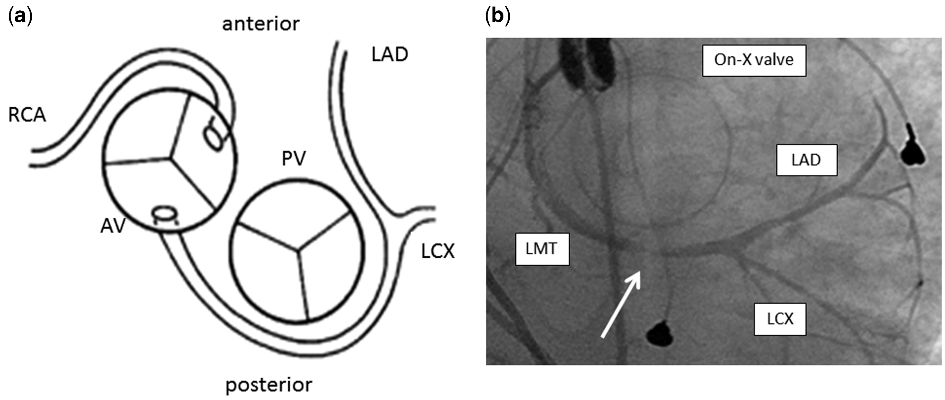

Figure 1. The coronary artery in this case. (*a*) Schematic view of Shaher type 9; the LMT runs along the back of the pulmonary valve. (*b*) Coronary angiography revealed 90% stenosis of the LMT. The white arrow indicates stenosis. AV = aortic valve, LAD = left anterior descending, LCX = left circumflex, LMT = left main trunk, PV = pulmonary valve, RCA = right coronary artery.

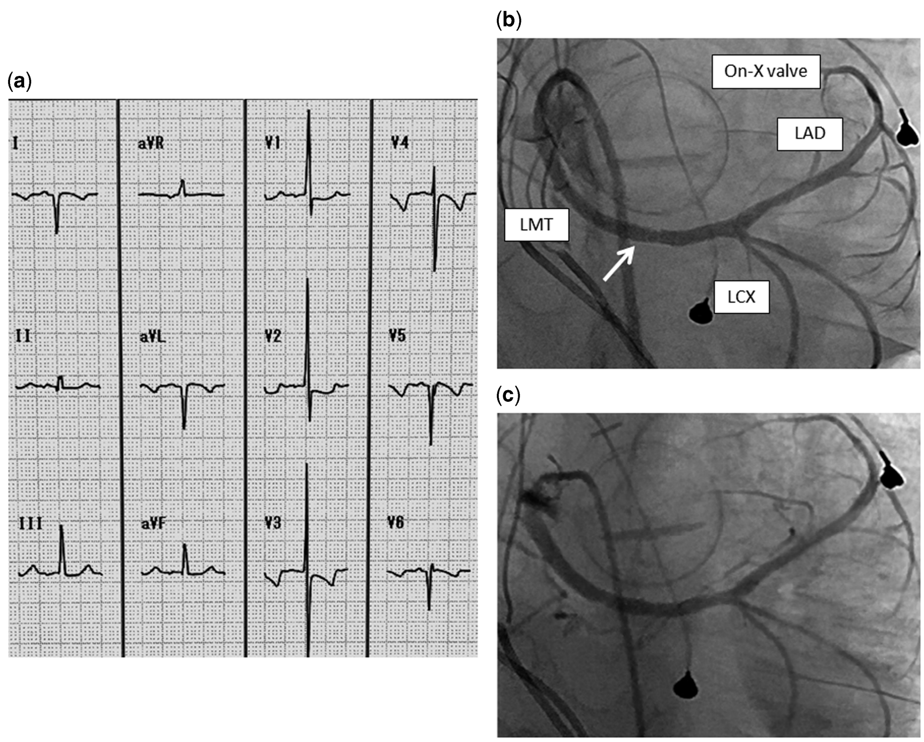

Figure 2. ECG and coronary angiogram after AVR. (*a*) ECG showed ST-T alteration and Q wave in the precordial leads. (*b*) Stent was sufficiently dilated in good position. The white arrow indicated the restoration of blood flow. (*c*) Angiogram at 4 months after stent deployment revealed complete stent patency. AVR = aortic valve replacement, ECG = electrocardiogram, LAD = left anterior descending, LCX = left circumflex, LMT = left main trunk.

After this procedure, dual antiplatelet therapy with aspirin and cilostazol was started. Follow-up angiography was performed at 4 months after stent implantation. All angiograms revealed complete stent patency (Fig 2c) and the ejection fraction by Simpson method using transthoracic echocardiography improved to 34%.

Discussion

We could successfully perform coronary stent deployment into the anomalous coronary artery using the caudal view in complex CHDs.

Norwood operation is usually performed to make a new aortic root using the pulmonary artery for the hypoplastic aorta. In this operation, the pulmonary valve is used as the neo-aortic valve. After this operation, neo-aortic regurgitation is a concern because exposure to systemic blood pressure or annular distortion causes functional deterioration in the pulmonary valve. Mechanical valve replacement is occasionally required,[3] but carries the risk of coronary artery compression because of abnormal anatomic spatial arrangement.[4] We considered reimplantation of smaller mechanical valve; however, previous reports have recommended an average diameter of 23 mm in children and showed the risk of aortic valve replacement under low cardiac function with coronary stenosis.[5,6] We performed coronary stent implantation for the left main trunk stenosis instead of reimplantation of smaller mechanical valve.

https://doi.org/10.1017/S1047951118002512 Published online by Cambridge University Press

Stent deployment in children mainly poses the risk of restenosis and acute thrombotic stent occlusion. However, coronary stents have been innovated from bare metal stents to drug-eluting stents, improving the risk of stent thrombosis.[7] Furthermore, Müller et al showed that the serum everolimus level after everolimus-eluting stent deployment is far below the immunosuppressive therapeutic range in infants.[8] The results of these past reports persuaded us to select everolimus-eluting stent.

Dual antiplatelet therapy using aspirin and clopidogrel or cilostazol after drug-eluting stent deployment is not only a well-recognized supportive care in adults but also effective in reducing the probability of thrombotic events and the restenosis rate in small children.[9] This patient had been taking warfarin and aspirin to prevent mechanical valve thrombosis. We added cilostazol to prevent restenosis, and follow-up angiography showed complete stent patency. In addition, Bratincsák et al reported that maintaining dual antiplatelet therapy for at least 12 months is crucial in infants after coronary stenting unless contraindications exist.[10] We plan to continue this treatment for at least 12 months.

These findings suggest that everolimus-eluting stent implantation is useful in rescuing coronary stenosis in small children with anomalous coronary artery.

Acknowledgements. We thank Dr Naoei Isomura for her help with the coronary intervention and helpful insights.

Financial support. This research received no specific grant from any funding agency, or from commercial or not-for-profit sectors.

Conflicts of interest. None.

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