Coronary artery ostioplasty without a patch for congenital left main coronary artery ostial atresia

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Video clip is available online.

Congenital left main coronary artery ostial atresia (LMCAOA) is an extremely rare coronary anomaly, and, thus far, there are only approximately 100 cases reported in the literature. Patch ostioplasty is gradually becoming the major surgical technique to treat LMCAOA, rather than coronary artery bypass graft. Ostioplasty without a patch has never been reported in the literature, with the exception of 1 case of ostial dilatation. However, the patient in that case did not survive the surgery, and the details of the procedure were not described. We describe the case of a 6-month-old infant with LMCAOA who underwent a successful ostioplasty without a patch and with no adverse postoperative events. The institutional review board/ethics review board and date of approval are XHEC-D-2022-145 and July 20th, 2022, respectively. Written consent was obtained from the parents.

CASE REPORT

A 6-month-old male infant who was asymptomatic and in excellent general condition after birth had a sudden worsening of his condition over 4 weeks following an episode of pneumonia. On examination, the heart sounds were distant, and a grade II/VI systolic murmur was heard in the mitral area. Electrocardiography revealed sinus rhythm without the typical indicators of myocardial ischemia. Transthoracic echocardiography showed that the origin of the left main coronary artery (LMCA) was indistinct, with retrograde blood flow in the left anterior descending branch (Figure 1, A and B), combined with severe dilatation of the left ventricle and moderate mitral valve regurgitation. Therefore, an anomalous origin of the left coronary artery was suspected, and a diagnosis of anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) was considered. Cardiac computed tomography showed a possible connection of the LMCA to the distal left posterior wall of the main pulmonary artery, supporting the diagnosis of ALCAPA (Figure 1, C and D).

As angiocardiology is not routinely performed for ALCAPA at our center, the patient underwent immediate surgical treatment. The pulmonary artery was opened; however, no coronary artery ostium was identified. A transverse aortotomy was then performed, and exploration revealed a normal ostium of the right coronary artery, with no ostium of the LMCA. A tenuous LMCA was observed after transection of the main pulmonary artery. The LMCA showed a

CENTRAL MESSAGE

For congenital left main coronary artery ostial atresia combined with a dysplastic left main coronary artery stem, ostioplasty without a patch might be a feasible method.
normal distribution and was connected to the lateral wall of the aorta (Figure 1, E). The surgeon attempted insertion of a 24-G puncture needle into the lumen of the LMCA from inside the aorta but failed (Figure 1, F). The anterior wall of the LMCA was incised, and the incision was extended into the aortic lumen to form an ostium. The luminal diameter of the LMCA was approximately 1 mm. The incision was then closed with 4 interrupted sutures (Figure 2). Directly after closure of the aorta and pulmonary artery, the heart activity was restarted, exhibiting forceful contraction and a normal electrocardiogram (Video 1). The patient’s postoperative period was uneventful. At the last follow-up (5 months after the operation), the patient was in good general condition. Echocardiography and computed tomography scan showed that the ostium of the LMCA was patent, and the LMCA trunk was well filled. The left ventricular systolic dysfunction and mitral valve regurgitation had decreased significantly (Figure E1).

**FIGURE 1.** LMCAOA displayed on echocardiography, CT, and during operation. A, The ostium and distribution of the LMCA are indistinct on echocardiography. B, Only retrograde blood flow is observed in the LMCA. C, Preoperative CT shows no origin of the LMCA from the aorta. D, The LMCA appears to connect with the pulmonary artery. E, The LMCA and its branches are dysplastic. F, The surgeon attempts insertion of a puncture needle into the lumen of the LMCA from inside the aorta but fails. The yellow arrow indicates the LMCA. LMCAOA, Left main coronary artery ostial atresia, CT, computed tomography; LMCA, left main coronary artery.
DISCUSSION

Congenital LMCAOA is an extremely rare coronary anomaly, and this case was the first in our center. In recent years, coronary artery ostioplasty has become the predominant procedure for treating LMCAOA.4

Because coronary angiography is not routinely performed for ALCAPA in our center due to radiation hazards and economic limitations, LMCAOA was confirmed during surgery in this patient. The surgeon first performed an ostioplasty as described previously, and coronary artery bypass graft was planned if the ostioplasty failed. Fortunately, ostioplasty was successful, and the patient recovered well after the operation.

Although the ostium and LMCA are only approximately 2 mm in diameter postoperatively, the patient is in good condition, indicating sufficient blood flow through the LMCA. This patient’s outcome indicates that we may underestimate the growth potential of the LMCA, which may be considerable after reconnection to the aorta. However, long-term follow-up is necessary to evaluate the risk of restenosis.

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FIGURE E1. Improved LMCA and MR. A, Antegrade flow in the LMCA 1 week after the operation. B, Antegrade flow in the LMCA 4 months after the operation. C, The LMCA is 0.181, 0.188, and 0.159 cm at the ostium, median, and distal parts, respectively, 4 months after the operation. D, CT image showing the LMCA connecting with the aorta before patient discharge. E, Enlarged left ventricle and significant mitral valve regurgitation preoperatively. F, Significant reduction in left ventricular dilatation and mitral valve regurgitation. The yellow arrow indicates LMCA (A-D) or MR (E-F). LMCA, Left main coronary artery; MR, mitral valve regurgitation; CT, computed tomography.