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Case Report Open Access

Left main coronary artery osteoplasty with pulmonary autograft in a child with familial hypercholesterolemia

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ABSTRACT

Familial hypercholesterolemia is a disease characterized by mutations in the low-density lipoprotein receptor. Total cholesterol levels >500 mg/dL and low-density lipoprotein levels >350 mg/dL are associated with early atherosclerosis. In these patients, the incidence of coronary artery disease is high in the first decade of life, and it often emerges in adolescence. Herein, we present a 14-year-old female with familial hypercholesterolemia who underwent left main coronary artery osteoplasty with a pulmonary autograft due to an incidentally detected osteal stenosis of the left main coronary artery. The patient was discharged without any problems, and no problems were detected during the follow-up.

Keywords: Familial hypercholesterolemia, left main coronary osteal stenosis, pulmonary autograft.

Coronary artery disease in childhood is rarely observed. Today, advances in detecting the presence of myocardial ischemia have led to the expansion of coronary revascularization indications in childhood. The most common cause is Kawasaki disease and there are few studies on other causes.[1] These include the ALCAPA (abnormal origin of the left coronary artery from the pulmonary artery) syndrome, which is not suitable for the reimplantation technique; left main coronary artery (LMCA) atresia, iatrogenic injuries and complications, and premature atherosclerosis, particularly during surgeries requiring coronary manipulation, such as arterial switch operations and Ross operations.[2] Coronary artery disease is observed in 10% of patients with familial hypercholesterolemia (FH), and coronary revascularization has been reported in the literature only as a case report.[3] Here, we present a patient with FH who underwent osteoplasty with a pulmonary autograft due to LMCA osteal stenosis.

CASE REPORT

A 14-year-old girl was scheduled for liver transplantation due to FH. The patient was referred to our clinic for preoperative evaluation. On physical examination, widespread xanthomas were observed in the joint regions. Heartbeats were rhythmic and normal. Electrocardiography was normal, and echocardiography demonstrated left ventricular hypertrophy and mild aortic regurgitation. Coronary angiography was planned for the patient, who had a history of rapid fatigue and exertional dyspnea and a family history of early death due to coronary artery disease. Angiography revealed LMCA osteal stenosis (Figures 1). The decision to operate was made with consensus. After median sternotomy, bicaval cardiopulmonary bypass was established in mild hypothermia (32°C), and cardiac arrest was achieved by del Nido cardioplegia (homemade). Myocardial protection was performed with retrograde cardioplegia. The ascending aorta and the pulmonary artery trunk were separated, and the LMCA was identified. An oblique incision was initiated on the anterior aspect of the aorta above the commissures and was extended across to the left lateral wall toward the LMCA. The anterior aspect of the LMCA was

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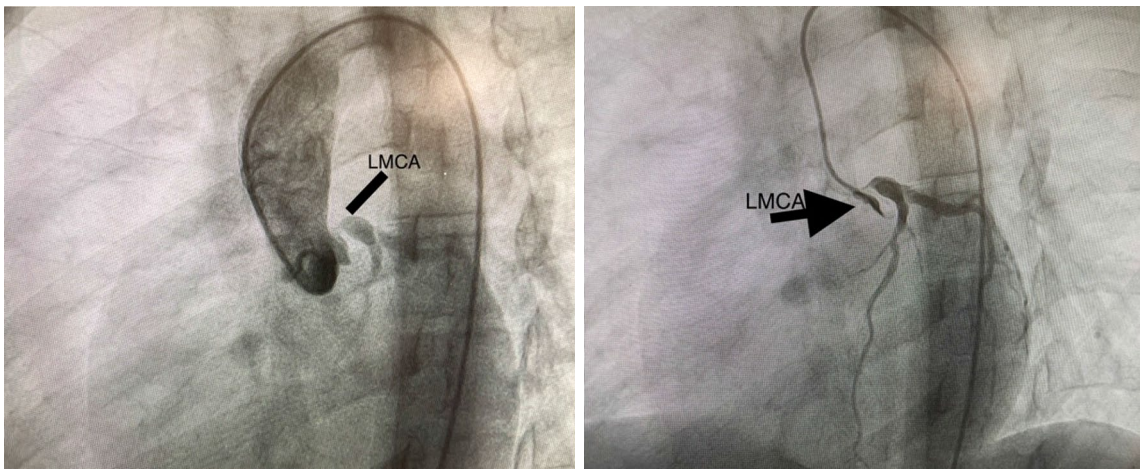

Figure 1. Preoperative angiographic images.
LMCA: Left main coronary artery.

opened for approximately 1 cm beyond the ostial lesion. During surgery, it was observed that the patient's aortic wall was extremely thick and plated. The patch was prepared from a fresh pulmonary artery. Left main coronary artery osteoplasty was performed with the anterior transaortic approach using the pulmonary autograft. The pulmonary artery defect was reconstructed with an autologous pericardial patch. The surgery was successfully finished. The patient was discharged without an issue on the seventh postoperative day, and no pathology was observed in the follow-up angiography at six months (Figure 2). The patient underwent a successful liver transplant eight months postoperatively.

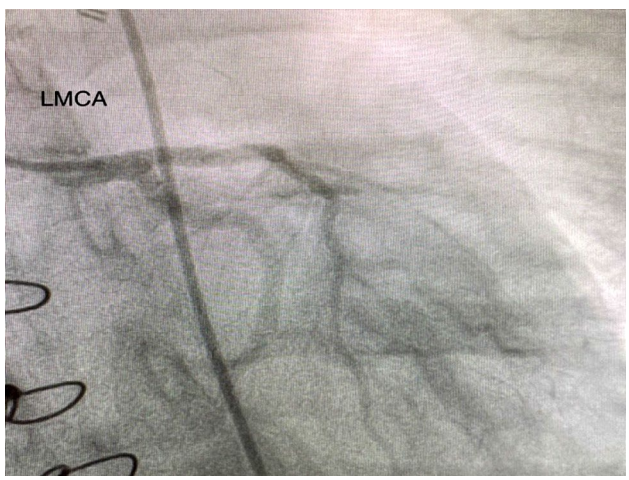

Figure 2. Postoperative angiographic image.

DISCUSSION

Guidelines for surgical indications for children with coronary artery disease have been developed in Japan. Indications for pediatric coronary revascularization are the same for children as for adult cases and include ischemic signs and symptoms. Unlike adults, however, children can be asymptomatic until the late term. Specifically, surgery is indicated if any of the following four conditions are present: *(i)* LMCA lesions, *(ii)* stenotic lesions of the multiple proximal coronary, *(iii)* proximal left anterior descending artery lesion, and *(iv)* collateral development.[4] Although our patient was asymptomatic, surgery was performed due to the presence of a severe proximal LMCA lesion.

Coronary artery surgery for children should be handled differently than that for adults as it involves some technical difficulties. These include the size of the coronary arteries, the difficulty of exposure, and the accessibility of the appropriate graft. Another problem is the long-term patency of the graft as children develop rapidly and their ability to lead a normal life depends on it. In 1966, Cooley et al.[5] first reported revascularization with an autologous saphenous vein graft in a baby with an abnormal left coronary artery. Later, with the use of the internal mammary artery and its long-term patency and growth potential, arterial grafts were preferred.[6] The patency of arterial grafts has been shown to be 3.5 times higher than that of venous grafts, even in children younger than three years old. Yatsunami et al.[7] reported that even in the majority of the neonatal population, the

coronary artery diameter is greater than 1 mm and the use of arterial grafts is appropriate.

The surgical technique to be applied in cases of proximal LMCA stenosis is controversial. The traditional surgical treatment for isolated LMCA osteal stenosis is coronary artery bypass grafting (CABG). Although this approach is effective, CABG can cause competition and steal phenomena. In addition, in cases of isolated osteal stenoses, retrograde perfusion to a large myocardial area is provided only with the graft.[8] Mavroudis et al.[1] stated that the combination of LMCA-plasty and CABG can be effective in this patient group. However, in the follow-up of their patients, string-sign findings were observed in the grafts in almost half of the cases. It was reported that only osteoplasty is sufficient for these patients. We preferred LMCA osteoplasty instead of CABG since our patient had a history of FH and would need to use long-term immunosuppressants after liver transplantation.

Although surgical osteoplasty was defined for adult patients by Effler et al.[9] and Sabiston et al.[10] in 1965, it was not preferred due to high mortality rates. Hitchcock et al.[11] obtained good results in 1983 with a posterior approach and Dion et al.[12] with an anterior approach in 1997. Finally, Liska et al.[13] suggested the transaortic approach, in which the aorta is transected and anteriorly mobilized to better visualize the coronary ostium and its distribution and facilitate patch reconstruction. We also prefer the transaortic approach during osteoplasty in our clinic. It allows the lesion to be better visualized, and it provides convenience while suturing the patch during repair.

The biggest advantages of surgical angioplasty in the pediatric population are that it does not require bypass material and provides antegrade flow. It has been used in cases of LMCA atresia, occlusion, and inflammatory arteritis.[14] However, there are few studies on proximal coronary artery patch-plasty. Prêtre and Turina[15] reported successful results in patients with LMCA osteal stenosis after surgical angioplasty. Successful reconstruction of both LMCA and RCA osteal stenosis with an autologous pericardium prepared in a "pantaloon" shape was also performed in another study.[16] This technique achieved good results in some selected cases of atherosclerotic lesions.[17] The success rate is high for lesions in the proximal half of the isolated noncalcified coronary trunk. However, an increased risk of thrombosis has been demonstrated for patients requiring extension of osteoplasty to the LMCA branches.[18] The use of autologous pericardium also has risks of fibrotic thickening, contraction, calcification, and late aneurysm formation due to exposure to systemic pressure. Its advantages are that it is flexible, resistant to infection, and nonimmunogenic.[19] Another material used in LMCA osteoplasty is the autologous pulmonary artery. It contains the intima, media, and adventitia layer. Long-term results of arch constructions with autologous pulmonary artery patches have been reported.[20] However, there are no definitive results about their use in osteoplasty in the literature. Ma et al.[21] reported positive early-and mid-term outcomes after LMCA-plasty. Ischemia was not observed in any of their patients. The pulmonary autograft had advantages due to material thickness, ease of manipulation, genetic homogeneity, and potential for somatic growth. Moreover, there was no risk of ectasia seen with pericardial patches. The use of pulmonary artery patches is thought to be more appropriate in our clinic. Pulmonary artery defects are also reconstructed with autologous pericardium to prevent the risk of pulmonary stenosis.

In conclusion, although patients with FH are asymptomatic, the risk of early atherosclerosis should not be forgotten. In cases of isolated LMCA osteal stenoses, osteoplasty may be a good option. The use of pulmonary autografts may be advantageous.

Patient Consent for Publication: A written informed consent was obtained from the parent of the patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

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REFERENCES

1. Mavroudis C, Backer CL, Duffy CE, Pahl E, Wax DF. Pediatric coronary artery bypass for Kawasaki congenital,

post arterial switch, and iatrogenic lesions. Ann Thorac Surg 1999;68:506-12.
2. Thammineni K, Vinocur JM, Harvey B, Menk JS, Kelleman MS, Korakiti AM, et al. Outcomes after surgical coronary artery revascularisation in children with congenital heart disease. Heart 2018;104:1417-23.
3. Kitamura S. A new arena in cardiac surgery: Pediatric coronary artery bypass surgery. Proc Jpn Acad Ser B Phys Biol Sci 2018;94:1-19.
4. JCS Joint Working Group. Guidelines for diagnosis and management of cardiovascular sequelae in Kawasaki disease (JCS 2013). Digest version. Circ J 2014;78:2521-62.
5. Cooley DA, Hallman GL, Bloodwell RD. Definitive surgical treatment of anomalous origin of left coronary artery from pulmonary artery: Indications and results. J Thorac Cardiovasc Surg 1966;52:798-808.
6. Viola N, Alghamdi AA, Al-Radi OO, Coles JG, Van Arsdell GS, Caldarone CA. Midterm outcomes of myocardial revascularization in children. J Thorac Cardiovasc Surg 2010;139:333-8.
7. Yatsunami K, Nakazawa M, Seguchi M, Momma K, Imai Y. The size of the coronary arteries in children with complete transposition before and after the arterial switch operation. Cardiology in the Young 1994;4:340-6.
8. Hutter JA, Pasaoglu I, Williams BT. The incidence and management of coronary ostial stenosis. J Cardiovasc Surg (Torino) 1985;26:581-4.
9. Effler DB, Sones FM Jr, Favaloro R, Groves LK. Coronary endarterotomy with patch-graft reconstruction: Clinical experience with 34 cases. Ann Surg 1965;162:590-601.
10. Sabiston DC Jr, Ebert PA, Friesinger GC, Ross RS, Sinclair-Smith B. Proximal endarterectomy, arterial reconstruction for coronary occlusion at aortic origin. Arch Surg 1965;91:758-64.
11. Hitchcock JF, Robles de Medina EO, Jambroes G. Angioplasty of the left main coronary artery for isolated left main coronary artery disease. J Thorac Cardiovasc Surg 1983;85:880-4.
12. Dion R, Elias B, El Khoury G, Noirhomme P, Verhelst R, Hanet C. Surgical angioplasty of the left main coronary artery. Eur J Cardiothorac Surg 1997;11:857-64.
13. Liska J, Jönsson A, Lockowandt U, Herzfeld I, Gelinder S, Franco-Cereceda A. Arterial patch angioplasty for reconstruction of proximal coronary artery stenosis. Ann Thorac Surg 1999;68:2185-9.
14. Sepehripour AH, Harling L, Ashrafian H, Athanasiou T. Pediatric applications of surgical patch angioplasty of the main coronary trunks. World J Pediatr Congenit Heart Surg 2014;5:283-90.
15. Prêtre R, Turina MI. Surgical angioplasty of the left main coronary artery in non-atherosclerotic lesions. Heart 2000;83:91-3.
16. Prifti E, Bonacchi M, Luisi SV, Vanini V. Coronary revascularization after arterial switch operation. Eur J Cardiothorac Surg 2002;21:111-3.
17. Bonacchi M, Prifti E, Giunti G, Leacche M, Ballo E, Furci B, et al. Mid-term outcome of surgical coronary ostial plasty: our experience. J Card Surg 1999;14:294-300.
18. Meseguer J, Hurlé A, Fernández-Latorre F, Alonso S, Llamas P, Casillas JA. Left main coronary artery patch angioplasty: Midterm experience and follow-up with spiral computed tomography. Ann Thorac Surg 1998;65:1594-7.
19. Ohba M, Matsuo K, Kabasawa M, Kito H, Asano S, Hirano M, et al. Surgical angioplasty for ostial atresia of left main coronary artery in child. Gen Thorac Cardiovasc Surg 2015;63:239-41.
20. Roussin R, Belli E, Lacour-Gayet F, Godart F, Rey C, Bruniaux J, et al. Aortic arch reconstruction with pulmonary autograft patch aortoplasty. J Thorac Cardiovasc Surg 2002;123:443-8.
21. Ma K, Qi L, Yuan J, Zhang B, Yuan X, Fan T, et al. Anatomic repair of left main coronary artery atresia: Coronary ostioplasty with autologous pulmonary artery. Can J Cardiol 2021;37:887-94.