

Repair of anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) by sinus pouch technique

Masaru Kumae

Chiba University Hospital

Mitsuru Aoki (✉ m.aoki@outlook.com)

Chiba University Hospital

Ikuo Hagino

Chiba University Hospital

Hiroshi Koshiyama

Chiba University Hospital

Takahiro Ito

Chiba University Hospital

Hironobu Nishiori

Chiba University Hospital

Case Report

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Abstract

Background: ALCAPA is a rare disease. The sinus pouch technique reported in transposition of the great arteries has not been reported to be applied to ALCAPA, but it was a useful method.

Case presentation: A five-month-old girl, weighing 5.3 kg, diagnosed ALCAPA underwent emergency surgery. The left coronary artery (LCA) originated from the posterior pulmonary artery (PA), and the left main trunk (LMT) was very short (1.5 mm), with moderate level of mitral valve regurgitation (MR). Because of the low height from the orifice to the pulmonary valve (Pv), it was necessary to avoid the tunneling technique due to concerns about adverse effects on the Pv. The sinus pouch technique was chosen because of its simplicity and low risk of stenosis due to coronary artery bending, especially in cases with short LMT.

Conclusion: We report a case of infantile ALCAPA with good results using this technique.

Background

ALCAPA causes myocardial ischemia, which leads to impaired cardiac function and MR. Various methods of coronary artery grafting have been reported. Necessity of intervention for MR, however, has been discussed. To the best of our knowledge, there is no report of ALCAPA with coronary artery implantation by applying sinus pouch technique¹, which has been reported in transposition of the great arteries.

Case Presentation

A five-month-old girl weighing 5.3 kg with no significant medical history had been diagnosed after worsening of heart failure symptoms. Electrocardiogram revealed ST elevation at V1-2, negative T at V1, V2, V5-6. BNP was 3379 pg/ml. Echocardiography indicated LCA from right posterior sinus of the PA and short distance to Pv, left ventricular enlargement (left ventricular end-diastolic volume (LVEDV) was 493% of normal(%N))², Z-score was -6.25, decreased cardiac function (left ventricular ejection fraction (LVEF) was 15%), and moderate level of regurgitation from the anterior and posterior commissures of the mitral valve. Computed tomography (CT) enabled identification short LMT (1.5 mm), the distance from the LCA orifice to the aorta is too far (Fig.1 pre-operative CT). LCA implantation by sinus pouch technique and mitral annuloplasty were scheduled on an emergency basis.

Under cardiopulmonary bypass, PA was transected at the bifurcation. The coronary orifice was found in the right facing sinus near the left posterior commissure (Fig.2-A Surgical observations(schema)-A). The coronary orifice was detached with a large cuff of two posterior Valsalva sinuses (Fig.2-B). By suturing the right and left parts of the Valsalva sinus flap, the LMT extension was created (Fig.2-C). An inverted L-shaped incision was placed in the ascending aorta and the coronary tube was anastomosed (Fig.2-D). The defect in the posterior sinuses of the PA trunk was supplemented with autologous pericardial patch. Kay-Reed annuloplasty was placed at the anterolateral and posteromedial commissures of mitral valve.

We considered using Extracorporeal Membrane Oxygenation (ECMO), but she was able to wean from cardiopulmonary bypass. On postoperative day three, delayed sternal closure was performed. She was discharged after adjusted heart failure medications. At the time of discharge, her cardiac function had not improved significantly and was equivalent to the preoperative level. There was no worsening of MR.

Eight months after the surgery, CT indicated a smooth coronary route with no obstructions (Fig.3 post-operative CT). Electrocardiogram revealed without ST-T changes. BNP was 49 pg/ml. One year after the surgery, echocardiography showed improvement of left ventricular enlargement (LVEDV was 17 %N) and cardiac function (LVEF was 6 %). MR was residual mild-moderate levels from the anterolateral commissure. Catheterization indicated a smooth coronary route with no obstructions, LVEDV was 195%N, LVEF was 71%, MR was Sellers II'.

Discussion And Conclusions

It is said that transplantation is possible in all cases³, and the 2-coronary system is the basic approach. The method of coronary artery reconstruction should take account of various factors such as location of the coronary artery orifice, distance from the aorta, and length of the LMT. In cases where direct grafting is not feasible, LMT extension formation should be considered in cases where the orifice is close to the Pv, where the tunnel technique may have an adverse effect on the Pv. There have been several reports of creating a tube with a PA wall flap where direct grafting is difficult. This technique, which creates a longitudinal flap against the Pv and utilizes the sinus bulge, is advantageous in that it does not involve movement of the coronary artery itself and requires less dissection, thus avoiding stenosis, and it reduces the risk of bleeding by a shorter suture line compared to other LMT extension methods. However, there may be a risk of thrombosis due to the swelling of the LMT, and attention should be paid to anticoagulation and follow-up.

The causes of MR have been reported to include papillary muscle dysfunction by ischemia and annular enlargement caused by left ventricular dilation⁴. Opinions are divided as to intervene in MR or not. The aggressive groups have suggested that intervention for MR should be based on the following reasons: simple methods such as Kay-Reed are expected to improve postoperatively, aortic cross clamp time should not be prolonged excessively, and in cases other than infants, irreversible abnormalities of the papillary muscle and tendons have occurred and improvement is not expected without intervention⁴. On the other hand, the conservative groups argue that mitral valve intervention should not be performed at the same time as coronary artery transplantation for the following reasons: spontaneous improvement can be expected in the remote period, the valvuloplasty increases the aortic cross clamp time, regurgitation is difficult to control in ischemic MR based on adult experience, and regurgitation control would lead to an increase in postoperative afterload, etc⁵. We have a policy of performing at least a valvuloplasty such as the Kay-Reed technique in patients with more than moderate regurgitation on preoperative echocardiography, and if the mitral valve is checked intraoperatively and structural abnormalities are found, we will actively intervene simultaneously in the valve and subvalvular area. Although initial valvuloplasty may not be sufficient to control regurgitation in patients with structural

abnormalities, but it is expected to be effective in preventing further exacerbation of regurgitation. If the aortic cross clamp time is prolonged, consideration should be given to admission to the ICU with planned use of ECMO. To our knowledge, this is the first report of infant type ALCAPA to use sinus pouch technique with good prognosis.

Declarations

Ethics approval and consent to participate: Not applicable

Consent for publication: Consent is obtained from the patient's parents.

Availability of data and materials: The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Competing interests: The authors declare that they have no competing interests

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Figures

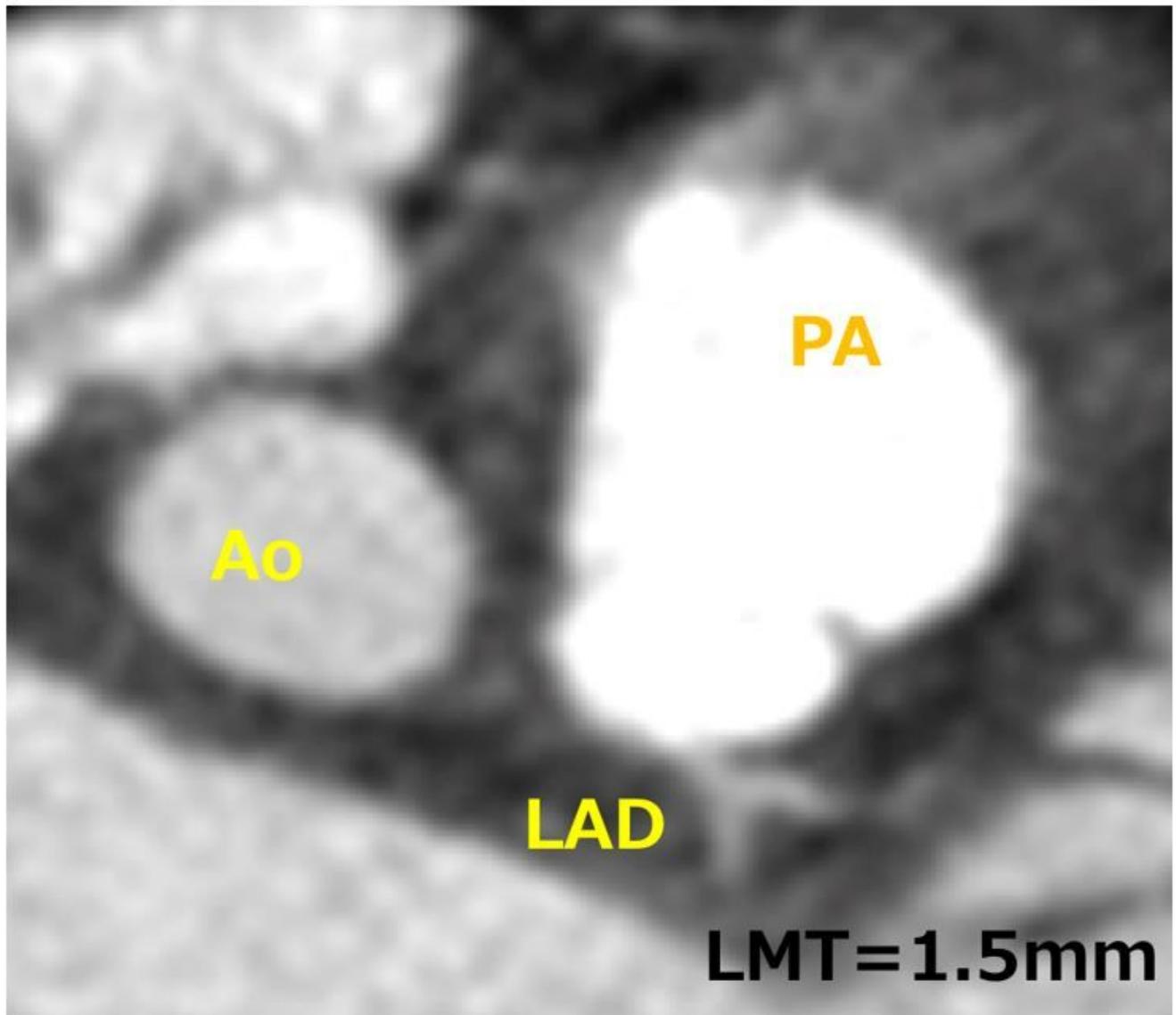


Figure 1

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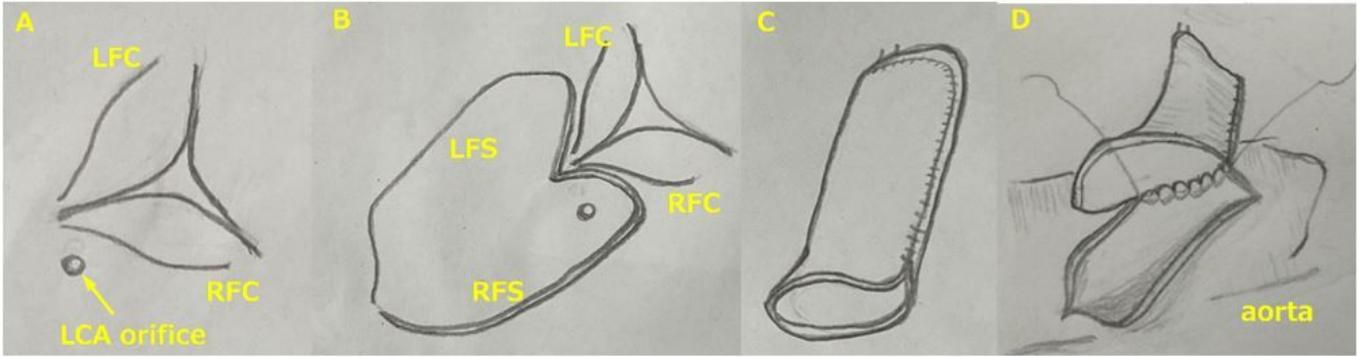


Figure 2

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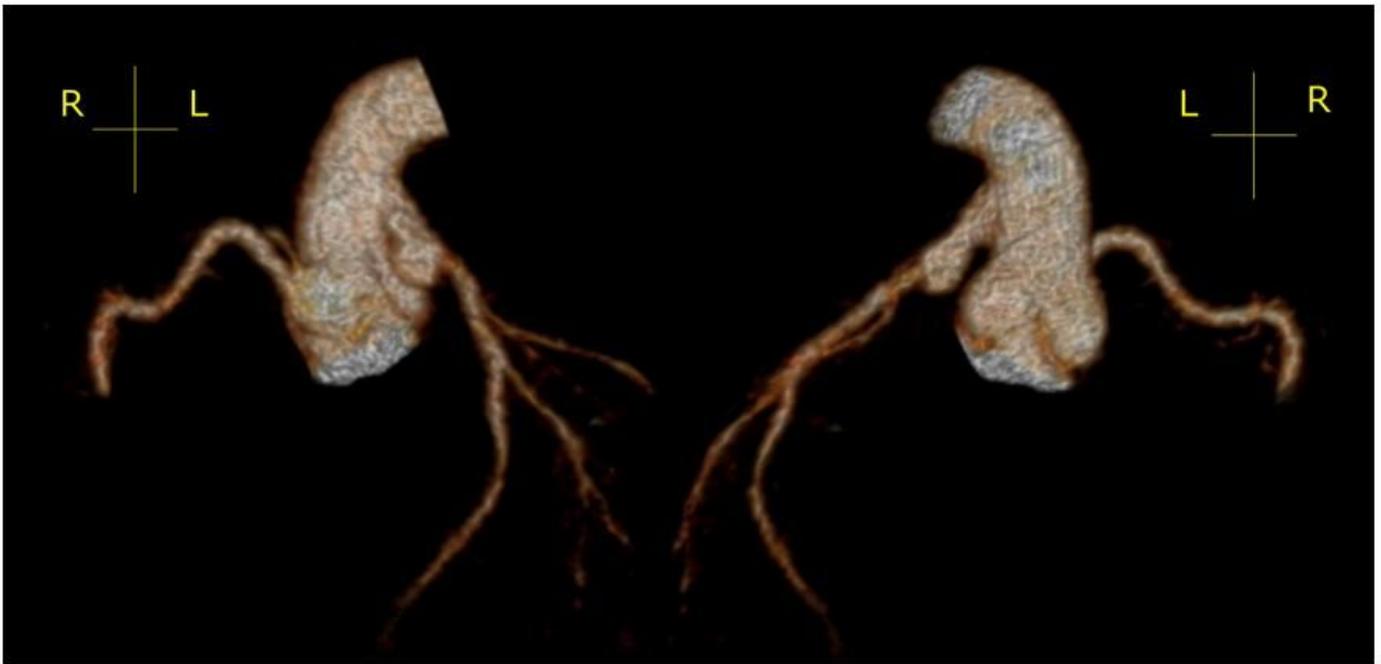


Figure 3

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