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Percutaneous coronary intervention in a 4-month-old infant for acute myocardial ischemia after repaired anomalous left coronary artery from the pulmonary artery

Mirsad Kacila, Mirza Halimić*, Merjema Karavdić, Almira Kadić, Saša Lukić, Sanko Pandur, Nusreta Hadžimuratović

Specialized Hospital "Heart Center", Sarajevo, Bosnia and Herzegovina

ABSTRACT

Anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) is rare, but life-threatening condition. The treatment of choice in patients with ALCAPA is the establishment of a dual coronary artery system with surgical reimplantation of the left coronary artery in the left coronary sinus. Percutaneous coronary intervention is infrequent in the pediatric population but can be a life-saving by promptly restoring flow to an obstructed coronary artery. It is a highly demanding and high-risk procedure in infants due to the technical difficulties and the small coronary artery diameter in infants.

Key words: Anomalous origin; left coronary artery; pulmonary artery; percutaneous coronary intervention; infant; heart failure; treatment

INTRODUCTION

Anomalies of the coronary arteries can be found in about 1% of the general population. Anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA), also known as Bland-White-Garland syndrome, is rare, but life-threatening condition accounts for approximately 0.5% of congenital heart defects and usually gives symptoms early (3-6 months of age). The treatment of choice in patients with ALCAPA is the establishment of a dual coronary artery system with surgical reimplantation of the left coronary artery in the left coronary sinus. Percutaneous coronary artery stent angioplasty is infrequent in the pediatric population but can be a life-saving by promptly restoring flow to an obstructed coronary artery. In technical sense, it is a highly demanding and high-risk procedure in infants and additionally limited by the need for eventual surgical intervention. The small coronary artery diameter in infants contributes to the complexity of percutaneous coronary intervention (PCI) procedures performed in this age group and present a host of periprocedural complications associated with catheter and stent to patient size mismatch: Occlusion ischemia, vessel dissection or rupture, and intimal flaps which can result in myocardial ischemia and infarction (1-3).

*Corresponding author: Mirza Halimić, Specialized Hospital "Heart Center," Sarajevo, Bosnia and Herzegovina.
E-mail: halimicm@gmail.com

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UNIVERSITY OF SARAJEVO
FACULTY OF HEALTH STUDIES

CASE REPORT

Male patient, 28 months old, was admitted to our hospital for an elective heart catheterization to check the possibility of the left main coronary artery (LMCA). At the age of 4 months, the patient was diagnosed with ALCAPA when surgical correction followed by stent placement in LMCA was performed.

Twenty-eight months after surgical correction and stent placement, heart catheterization was performed and coronary artery angiogram showed normalization of the left coronary vasculature and no signs of in-stent stenosis (Figure 1). After the catheterization, the patient was discharged home on salicylate and antihypertensive therapy. Clopidogrel was excluded because it was believed that its therapeutic activity has ended.

Before the diagnosis of ALCAPA, the baby was admitted to the hospital with the aspects of congestive heart failure, dyspneic breaths, and problems with feeding. Systolic murmur was distinguished. On electrocardiogram, signs of anterolateral myocardial infarction were noted. Dilatative cardiomyopathy with fractional shortening of 13% was noted on echocardiogram. The right coronary artery (RCA) originated from the aorta. The left coronary artery could not be envisioned originating from the aortic root. Suspicion on ALCAPA was brought and heart catheterization was indicated. Coronary angiogram showed anomalous left coronary artery originating from the posterior wall of the pulmonary artery. Surgery was indicated.

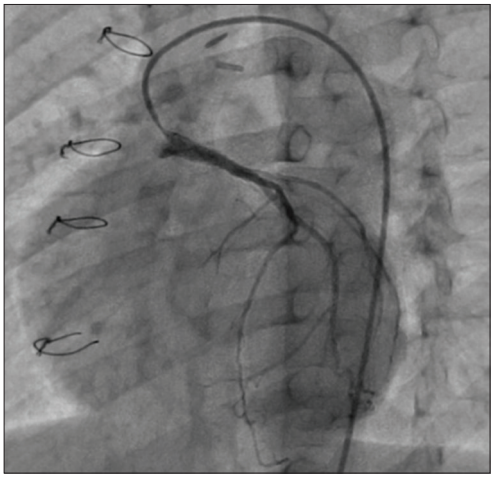

FIGURE 1. Coronary angiogram shows the left coronary artery 28 months after the reimplantation of anomalous origin of the left coronary artery from the pulmonary artery to aortic root and stent placement in the left main coronary artery. The complete resolution of flow in the left coronary artery is accomplished and no signs of in-stent stenosis are present.

The patient underwent reimplantation of ALCAPA to aortic root and was transferred to the intensive care unit on high inotropic support. Sternal closure was delayed and the sternum was closed on the 6th post-operative day. On the 6th post-operative day, *Enterobacter cloacae* was isolated from the tracheal aspirate. Regular echocardiograms revealed minimal improvement of overall myocardial improvement.

On the 8th post-operative day, the patient was extubated but had to be reintubated shortly after, due to the hemodynamic instability. In the following days, as myocardial dysfunction and dysrhythmia persisted, decision for coronary angiography was set. On the 13th post-operative day, catheterization was performed where coronary angiogram showed no antegrade flow in LMCA – aortic anastomosis with LMCA and the tube graft was obstructed. Circumflex artery and left anterior descending artery were getting supply by collateral arteries. Balloon angioplasty and stent implantation were performed inside the coronary tube graft which was created during the operation (4-4.5 mm in size): After one drug-eluting stent (XIENCE PRIME 3.5 mm × 8 mm) was implanted in LMCA, distal LMCA stenosis was absent on angiogram, but residual stenosis in the proximal portion of LMCA was present so that the second drug-eluting stent (XIENCE PRIME 4 mm × 9 mm) had to be implanted. LMCA flow presented to be normal after the procedure. After stent implantation, sinus rhythm persisted and regular echocardiograms revealed the improvement of the myocardial functions. Extubation followed on the 15th post-operative day, but reintubation had to be done on the next day due to respiratory distress and tracheostomy cannula was implanted.

The patient was transferred to the ward 27 days after the operation after removing tracheostomy cannula with improved myocardial and respiratory functions. The patient was discharged home 36 days after the operation on salicylate, clopidogrel, digoxin, diuretics, and ACEI therapy.

Two years after the procedure, the patient experienced few episodes of transient loss of consciousness but apart from that clinical condition was satisfying and stable.

The most recent echocardiogram performed as an outpatient 28 months after the procedure shows normal left ventricular (LV) function (fractional shortening – 35%; ejection fraction –66%). The latest electrocardiogram shows no changes within ST segment.

DISCUSSION

In infants, percutaneous coronary intervention is remained rare procedure and is associated with the technical difficulties and risk from complications. Only four cases of PCI in ALCAPA patients were reported previously. Long-term results of PCI in surgically corrected ALCAPA patients are to a great extent unknown. Two out of four reported cases had no restrictions in follow-up examinations at age 6 and 7 years, although one case required surgical reintervention for recurrent mitral regurgitation. The other two only reported follow-up data within 1 year post-PCI (4-7).

There are a limited data on drug-eluting stents in pediatric population (8,9). In a reported patient, it was elected to implant drug-eluting stents, with diameters ranging from 3.5 to 4 mm.

Dual antiplatelet therapy (salicylate and clopidogrel), a standard in adults following coronary stent implantation (10,11), proved to be essential in pediatric population to prevent in-stent restenosis and reduce morbidity and mortality.

Available literature presents two case reports regarding PCI in infants (4,12). The first report describes a 3-month-old infant after surgical repair of ALCAPA, who developed poor ventricular function due to LMCA obstruction, underwent PCI with a 2.25 mm diameter bare-metal stent implantation. The patient was discharged with single antiplatelet therapy – salicylate only. Complete resaturation of the LV ejection fraction was noted with echocardiogram 2 and 6 months after contrast score index (CSI) (4). The second case report described a 7-month-old infant with surgical repair of ALCAPA who developed complete obstruction of the LMCA and underwent balloon recanalization followed by stent scaffolding of the LMCA using a 2.5 mm diameter bare-metal stent. The patient was given dual antiplatelet therapy with salicylate and clopidogrel. Nevertheless, 3 weeks later, the child developed acute myocardial infarction due to 3 days of interruption in antiplatelet therapy secondary to gastrointestinal bleeding. PCI was repeated and the stent was dilated up to 3 mm. Due to persistent myocardial dysfunction, experimental local infusion of bone marrow-derived stem cells was performed. Follow-up echocardiogram after 14 months of CSI showed improved LVEF (12).

As in the previous two case reports, it is advised that PCI is a relatively safe procedure in infants and young children with congenital and acquired coronary artery stenosis. Coronary stent implantation can save a patient's life and improve the blood supply of myocardial infants in critical condition. Bypass surgery can carry a greater risk in children with damaged myocardial function or severe comorbidities when compared to cardiac catheterization.

However, percutaneous stent implantation is not meant to serve as a regular and definitive treatment in children <15 months of age. The coronary artery diameter is so small in young children that they could not accommodate stents that could be dilated to final diameters generally acceptable for adults. The diameter of the RCA measures about 3 mm and the LMCA 4.5 mm in adults (13).

Small children with stents placed in LMCA should be followed by regular imaging studies by preference cardiac catheterization. Stents can additionally be dilated as the child and the coronary artery grows. As an alternative, if PCI cannot additionally improve coronary flow, surgical repair is an option. Having in mind, the limited potential of stents implanted in infants and young children is important in deciding whether PCI with CSI should be pursued. Even though the risk is lower than risk within surgery, it should be done only in urgent situations. Extracorporeal membrane oxygenation support should be readily available.

It is recommended to use the largest available stent diameter based on adjacent coronary diameter to prevent or reduce the extremely uncommon scenario of in-stent restenosis. In infants undergoing CSI, it is pivotal to maintain dual antiplatelet therapy with salicylate and clopidogrel for at least 12 months unless there is a contraindication.

CONCLUSION

Coronary artery stent placement is an attainable and relatively safe option in infants and young children with coronary artery stenosis in urgent situations. Support by aggressive dual antiplatelet therapy is of a crucial importance in reducing the risk of in-stent stenosis. In these cases, CSI is a feasible method because it allows us to avoid urgent, high-risk coronary surgery.

In our patient, who was 4 months old when he underwent PCI on LMCA due to a total occlusion following repaired ALCAPA, we report the complete recovery of ventricular function which persisted at 28-month follow-up and complete and flow normalization of the left coronary artery system.

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