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Brief Report

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Urgent aortic valve neocuspidization using the Ozaki procedure in an infant with native aortic valve endocarditis

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Abstract

Native valve aortic endocarditis is rarely seen in the paediatric population. Although, the firstline of treatment is medical, surgical intervention may be indicated in patients with unrepairable valvular and subvalvular disease. Recently, the aortic valve neocuspidization (AVNeo) procedure has gained popularity both in adult and children in whom other repair techniques are not feasible. In this case report, we present an urgent aortic valve replacement using the AVNeo technique in a critically ill infant with a small annulus, severe left ventricular outflow tract stenosis, and severe aortic regurgitation.

Native aortic valve endocarditis is a critical illness that has high mortality and morbidity. It rarely occurs in children when compared with adults.¹ Surgical intervention is indicated when the aortic valve is severely damaged or if the left ventricular outflow tract is critically obstructed.¹ The best option is to repair the valve but if this is not feasible, then the replacement of the valve with a pulmonary autograft (Ross operation) or aortic homograft might be the only solution in aortic valve endocarditis.² In recent years, the aortic valve neocuspidization (AVNeo) technique has been performed in patients with endocarditis and has promising early and mid-term successful results.³ In this case report, we present an urgent aortic valve replacement using the AVNeo technique in a critically ill infant with a small annulus, severe left ventricular outflow tract stenosis, and severe aortic regurgitation.

Case report

A 3-month-old, 4500 g male patient was consulted to the paediatric cardiovascular surgery department for a vegetative mass on the aortic valve leading to severe aortic insufficiency and stenosis. The medical history of the patient revealed that he had been admitted to the hospital with complaints of respiratory distress on the postnatal second day. On echocardiographic evaluation, severe aortic valvular stenosis with a bicuspid aortic valve was detected. For this reason, a balloon angioplasty procedure had been performed on the postnatal third day. The patient had been hospitalised in the intensive care unit for 25 days before being discharged. After discharge, he was followed closely. Two months later, the patient was brought to the hospital with complaints of runny nose, cough, and diarrhoea that had continued for 1 week. The evaluation revealed a hyperechogenic mass which was 4.5×6.5 mm in diameter detected on the aortic valve. The mass was thought to be a thrombus or vegetation secondary to infective endocarditis, and low molecular weight heparin with vancomycin-meropenem antibiotic treatment was empirically started after drawing blood cultures for preliminary diagnoses. The patient had no septic or thromboembolic complications. Candida Parapsilosis proliferation was documented in the blood culture, and antifungal treatment with caspofungin was added to the antibiotic treatment. During the follow-up period, a progressive enlargement $(9.7 \times 6.8 \text{ mm})$ and high mobility of the mass on the aortic valve gave rise to haemodynamic abnormalities and urgent surgical intervention was decided for the patient (Video 1).

Surgical procedure

Following median sternotomy, standard aortobicaval cannulation was performed. Oblique aortotomy was conducted after cardioplegic arrest. A large vegetative mass that had damaged the right coronary and left coronary cusps of the aortic valve and caused severe stenosis at the level of sinus valsalva was observed (Fig 1). Considering that the mass could not be isolated from the valves, it was resected together with both destructed aortic valves. It was confirmed that the infection had not extended to the aorticomitral continuity and involved the non-coronary cusp (Fig 2). We then decided to perform the Ozaki procedure. The autologous pericardium was harvested and treated with 0.6% glutaraldehyde solution for 2 minutes, followed by rinsing with



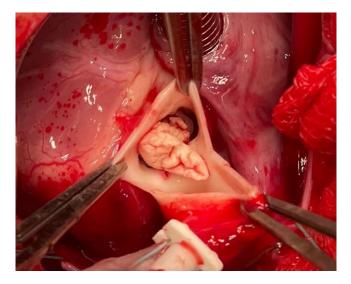


Figure 1. Intraoperative view of the vegetation

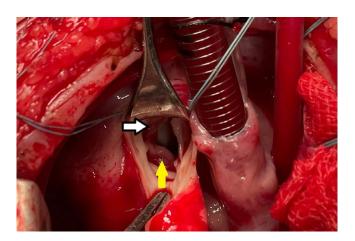


Figure 2. View of aortic cusps following removal of vegetation. White and yellow arrows indicate destructed right and left coronary cusps respectively.

saline solution three times for 6 minutes. Standard measurements were made for the removed valves using the Ozaki sizing apparatus. Then, the neocusps were cut for the left and right coronary cusps. The surgical procedure was performed as described in the literature.⁴ Cross clamp and cardiopulmonary bypass times were 62 and 86 minutes, respectively. Intraoperative epicardial echocardiography demonstrated correct movement of the neocusps and good coaptation with the native right coronary cusp. A maximum gradient of 20 mmHG was measured on the aortic valve with 1–2 degrees of aortic regurgitation. The patient was then transferred to the ICU with milrinone and dopamine infusions.

The patient was extubated on the post-operative second day. The subsequent echocardiographic study revealed a second degree eccentric aortic valve regurgitation and mild to moderate aortic valve stenosis (1.9 m/second) (Video 2). The patient was transferred to the ward with diuretic, ACE inhibitor, acetylsalicylic acid, and caspofungin treatments. The remaining postoperative course was uneventful, and the patient was discharged on the 20th postoperative day upon completion of the 4-week caspofungin treatment.

Discussion

AVNeo is an important treatment alternative that has gained popularity in recent years and has been used in various pathological conditions related to the aortic valve in both adult and children.^{2,5} With AVNeo surgery, one or more aortic valve leaflets can be replaced using autologous or heterologous pericardial tissue.⁶ Numerous studies, especially in the adult age group, have shown that this surgical method is associated with low mortality, morbidity, and reoperation in the early, medium, and long term.^{3,7} After the experiences in adult patients, the AVNeo technique has been frequently used in the childhood age group. Although the early and mid-term results are successful, data for long-term morbidity and reoperation rate have not yet been published.^{8,9}

A wide variety of surgical repair techniques are available for congenital or acquired aortic valve stenosis and insufficiency in children.² These techniques have acceptable results in the medium and long term.¹⁰ However, replacement is the only option for aortic valve pathologies that are unsuitable for surgical repair. There are various alternatives for aortic valve replacement in children, and these are pulmonary autograft implantation (Ross procedure), aortic homograft implantation, and mechanical valve replacement.¹¹ Although the Ross procedure has excellent results in the early and medium term, it has some drawbacks such as high mortality in the neonatal and infant period, the need for reoperation for root dilatation in the long term, and the need for long-term intervention for stenosis developing in valve conduits placed in the pulmonary position.¹² Homografts are reasonable alternatives for the aortic position in children. However, they are not the best option due to the lack of limitations in availability and a high rate of structural deterioration in the long term.¹³ Mechanical valves, on the other hand, have various disadvantages such as the life-long need for anticoagulation, risk of thromboembolism, possible need for additional aortic root expansion in patients with narrow annulus, and the need for reoperation due to pannus formation in the long term.14

The AVNeo technique has recently been used more frequently in children. The advantages of this surgical method can be listed as follows: it can be applied using autologous or xenografted pericardium; single or multiple valve leaflets can be replaced; it can be applied in isolation or using annulus expansion techniques; it allows the native aortic annulus to grow; it does not require lifelong warfarin anticoagulation due to the use of biological material; it offers an effective orifice area as good as an aortic bioprosthesis without a stent; and it has a low risk of thromboembolism. Thanks to these advantages, it seems to be suitable for children.

On the other hand, the AVNeo technique also has its drawbacks. First of all, a significantly long learning period may be required. Additionally, special sizing apparatus are needed for this technique to ensure valve competency. Since the surgical field in infants is inherently narrow, the reconstruction of the valve leaflets might be challenging in these patients and additional annular enlargement surgery might be required in some of them. Besides, the issues regarding the need for long-term anticoagulation and valvular durability have yet to be clarified.

In major studies recently published, we have seen that the AVneo technique has rarely been applied in the infant age group. This may be due to successful aortic valve repair techniques which provide enough time for patients to grow for a potential future Ross or valve replacement operation. Another possible reason may be that the surgeon has extensive experience with the Ross or Ross-Konno operation and also the lack of long-term results with AVNeo.

Two surgical options could have been performed on this patient. The first one was the Ross operation and the second, which was our choice, was the AVNeo surgery. The first reason we chose the AVNeo surgery was that the diameter of the pulmonary annulus was significantly larger than the diameter of the aortic annulus and an additional annular expansion operation would have been required for the patient. The second reason was due to the known disadvantages of the Ross operation. The third reason we favoured the AVNeo procedure was because the patient had severe heart failure requiring immediate intervention and was not in a good enough clinical condition to tolerate an extensive and lengthy operation such as the Ross procedure.

Although the experience in the adult and paediatric age groups has shown that AVNeo may be a safe alternative for aortic valve pathologies that are otherwise irreparable, we still have concerns about the durability of the replaced leaflets. It is evident that the calcium turnover is high in children and valve degenerations due to calcification are common when biological materials are used in valve repairs. This situation is more intense, especially in the systemic atrioventricular and semilunar valves. Since autologous pericardium treated with glutaraldehyde is used in the AVNeo technique, any potential degeneration risk is inevitable. That is why we decided to replace only the damaged valves rather than all three valves. We assumed that the more native leaflet tissue we preserve, the less calcification will develop. However, due to a lack of knowledge about the long-term valvular durability of the leaflets and the extent of calcific degeneration, we cannot say whether the original Ozaki procedure with three leaflet replacement is safer in the paediatric population or the limited leaflet replacement procedure that we conducted. We believe that all children who have undergone any type of AVneo surgery should be followed very closely.

To the best of our knowledge, this was the youngest infant to receive the AVNeo technique, who was not suitable for any other kind of repair due to the extensive destruction of the aortic valves. We believe that in case of urgency the AVNeo technique may be an alternative technique to Ross surgery in the infant age group with aortic valve endocarditis.

Supplementary material. To view supplementary material for this article, please visit https://doi.org/10.1017/S1047951122004267

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Conflicts of interest. None.

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