Postpericardiotomy Syndrome: A Rare Complication Following Double Arcus Aorta Operation

Postperikardiyotomi Sendromu: Çift Arkus Aort Ameliyatı Sonrası Nadir Bir Komplikasyon

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ABSTRACT

Vascular ring anomalies are rare congenital malformations and, a double aortic arch is the most common form. Correction rarely leads to postoperative complications. A full-term newborn was referred to our hospital on the postnatal 11th day due to periodic respiratory distress, feeding intolerance, and a diagnosesis of double arcus aorta. The case was operated on the postnatal 25th day and did not develop any problems. However, on the 8th day after the intervention, Postpericardiotomy Syndrome clinic occurred. Despite the fact that postpericardiotomy syndrome is common in children after undergoing open-heart surgery for congenital heart disease, to our knowledge, our case is the youngest in the literature to develop postpericardiotomy syndrome due to correction of the double aortic arch. In terms of understanding the reasons, we believe that this study may contribute to the literature.

Keywords: Newborn; Postpericardiotomy syndrome; Double aortic arch

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ÖZET

Vasküler halka anomalileri nadir görülen konjenital malformasyonlardır ve en sık görülen şekli çift aortik arktır. Zamanında doğan yenidoğan, periyodik solunum sıkıntısı, beslenme intoleransı ve çift arkus aort tanısı ile postnatal 11. günde hastanemize sevk edildi. Olgu postnatal 25. günde opere edildi ve herhangi bir sorun gelişmedi. Ancak girişim sonrası 8. günde Postperikardiyotomi Sendromu kliniği gelişti. Doğuştan kalp hastalığı nedeniyle açık kalp cerrahisi geçiren çocuklarda postperikardiyotomi sendromunun sık görülmesine rağmen bildiğimiz kadarıyla olgumuz çift aort arkının düzeltilmesine bağlı postperikardiyotomi sendromu gelişen literatürdeki en genç olgudur. Nedenlerinin anlaşılması açısından bu çalışmanın literatüre katkı sağlayacağı kanaatindeyiz.

Anahtar Sözcükler: Yenidoğan; Postperikardiyotomi Sendromu; Çift arkus aorta

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INTRODUCTION

Vascular ring (VR) is a condition that occurs as a result of compression on the trachea and/or esophagus due to the embryonic developmental disorder of the aorta and its main branches. Double aortic arch (DAA) and right aortic arch with aberrant left subclavian artery are the most common types of VR '(1)'.
Some complications such as phrenic nerve paralysis, recurrent laryngeal nerve paralysis, vocal cord paralysis, aortic esophageal fistula, esophageal erosion, and chylothorax may rarely develop after VR surgery '(2)'.
Postpericardiotomy syndrome (PPS) is a part of postcardiac injury syndrome and is defined as an inflammatory event that is developed due to the surgical intervention of the pericardium.
In this article, postpericardiotomy syndrome, which developed postoperatively in a newborn with a double aortic arch, was discussed due to its rare occurrence.

CASE REPORT

The full-term baby who had an uneventful delivery by vaginal route was referred to our hospital on postnatal 11th day due to periodic respiratory distress, feeding intolerance, and a diagnose of double arcus aorta. The parents had 3rd-degree consanguinity as first cousins and there was no family member with a similar history.

Computed tomography arteriography imaging revealed that the right arch aorta was present and was giving the right carotid and right subclavian artery branches; the left aortic arch was also seen, and it was found to give the left carotid and left subclavian artery branches (Figure 1). It was observed that there was compression on the esophagus at the carina level. The case was operated on the postnatal 25th day. Median sternotomy was performed and it was observed that the vascular ring was separated from the left subclavian artery and poured into the aorta from the posterior of the esophagus from the right course pattern. Meanwhile, the patent arterial duct was ligated and divided. The procedure did not involve cardiopulmonary bypass or pacemaker insertion. A total of 65 ml of hemorrhagic residual was detected from the thoracic drain on the first day of the patient who did not develop any other complications after the procedure. Control echocardiographic examinations were evaluated within normal limits.

The patient's stridor continued with a decrease in severity and fever above 38^{o}C developed on the 8th postoperative day. C-Reactive Protein: 16.1 mg\L (0-8 mg\L) , WBC: 21.8 10^3/µL, Hb; 12.4 g\dL, Platelet count; 457 10^3/µL were detected. Appropriate antibiotics were added to the treatment after receiving blood culture due to suspicion of infection. In the follow-up, respiratory distress and oxygen requirement increased gradually and echocardiographic analysis revealed an increase in pericardial effusion (Figure 2). Pericardiocentesis was performed. Approximately 40 ml of hemorrhagic fluid was drained by placing a pigtail catheter. Blood culture gave a negative result. Ibuprofen was given as an anti-inflammatory treatment. The patient was discharged on the 47th day of postnatal age, with recommendations to continue anti-inflammatory therapy. Informed consent was obtained from the legally authorized representative for publication of this case report and any accompanying images.

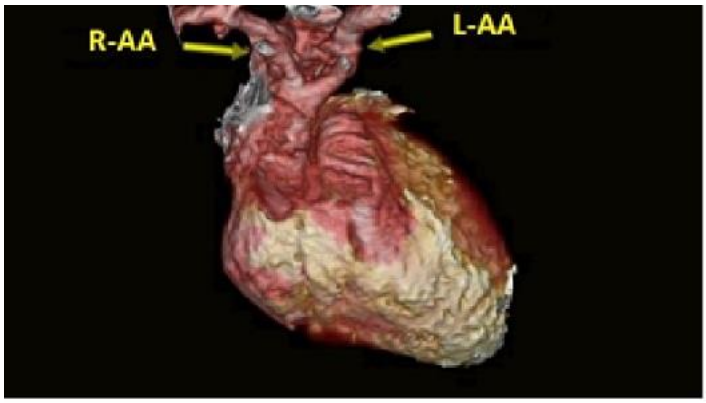


Figure 1. Computed tomography arteriography anterior view imaging of DAA.



Figure 2. Posteroanterior chest radiograph of patient after pericardiocentesis.

DISCUSSION

PPS refers to the clinical condition that develops after cardiothoracic surgery due to inflammation in the pleura, pericardium, or both. It is common in children undergoing open-heart surgery for congenital heart disease. Although, bleeding during surgery, coagulopathy, viral infections, and autoimmunity may also play a role '(3)'. Our case was operated on for DAA and PPS developed most probably due to intervention in the pericardium. Anticoagulant therapy was not given to the patient and laboratory tests did not reveal any coagulopathy or thrombocytopenia. The case did not have additional risk factors that could be explained in terms of the development of PPS. Moreover, the development of PPS in the early period of life decreases the possibility of autoimmunity.

PPS can be seen after pericardiotomy in all age groups and its incidence is thought to be between 1.6 and 3.5%. under 2 years of age '(4)'. Although cases with a diagnosis of PPS under 2 years of age have been described, especially after cardiac surgery, to our knowledge, our case is the youngest in the literature to develop the postpericardiotomy syndrome.

According to the latest recommendations of the European Society of Cardiology, ibuprofen is preferred as a first-line treatment because it has less side effect profile than alternative drugs in the treatment of PPS '(5)'. Since the patient was symptomatic and had excess pericardial effusion, pericardiocentesis was applied first, followed by ibuprofen treatment as an NSAID. It was decided to give ibuprofen treatment for 2 weeks.
Although the exact cause of PPS is still unknown, it is proposed that intervention in the pericardium appears to be the most important risk factor. During the surgical procedure of VR correction, especially if there is an intervention in the pericardium, close echocardiography should be followed in terms of the development of PPS and intensive care should be taken in terms of life-threatening complications.

Conflict of interest

No conflict of interest was declared by the authors.

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