Case Report

Surgical Treatment of Recurrent Tracheal and Esophageal Compression After Double Aortic Arch Surgery: Extra-Anatomic Bypass

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ABSTRACT

Vascular rings compress the trachea, the esophagus, or both, causing severe obstructive esophageal and airway symptoms. Its treatment is surgery. However, a second surgical intervention is required in patients with persistent symptoms after surgery. Sometimes, the reason for this may be that the initial surgery was insufficient. In the presented herein, we applied the extra-anatomic bypass and simultaneous ligamentum arteriosum division approach in a patient with a diagnosis of double aortic arch subjected to isolated arch division. ***(Iranian Heart Journal 2022; 23(2): 136-141)***

KEYWORDS: Reoperation, Vascular ring, Extra-anatomic bypass

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One of the rare causes of breathing and swallowing problems in childhood is the vascular ring. Vascular rings cause these symptoms by compressing the trachea and the esophagus. The vascular ring treatment is surgical, but persistent symptoms can be seen in the postoperative period. This situation can sometimes be related to inadequate surgical techniques. We herein describe a patient exhibiting persistent compression symptoms after double aortic arch (DAA) surgery.

Case Report

A 17-year-old male patient presented to the outpatient clinic with complaints of severe swallowing difficulty, intermittent dyspnea, feeling of choking while eating, and inability to gain weight. In the first computed tomography (CT) angiography of the patient, a double arch appearance was observed in the aortic arch. It was observed that the ring, formed by the aortic arch, completely surrounded and compressed the trachea and the esophagus. The right carotid and right subclavian arteries originated from the right aortic arch, and the left carotid and the left subclavian arteries originated from the left aortic arch (Fig. 1). The aorta descended from the right side of the vertebral column. The patient was diagnosed with DAA. Surgical correction was made for the patient in another hospital. Right anterolateral thoracotomy was performed. A division was applied to the segment where

the left (anterior) aortic arch joined the descending aorta.

The patient's symptoms partially resolved within a few months. However, there was never enough improvement. Complaints of dyspnea and vomiting while feeding started 3 years after the operation. The patient visited the emergency service many times because of dyspnea and started to lose weight. CT angiography performed on the patient 2 years after the first surgery showed that the right (posterior) aortic arch was intact. The right aortic arch was not connected to the descending aorta. Nonetheless, the right aortic arch and brachiocephalic artery exerted posterior pressure on the trachea and the esophagus (Fig. 2B). It was observed that the surgery performed on the patient could not sufficiently remove the compression caused by the ring. He was, therefore, scheduled to undergo surgery for a second time.

The patient was taken to the operating room. First, a sternotomy was performed in the supine position. The thymus was totally excised, and the pericardium was opened. Next, the left aortic arch and the right aortic arch were explored. The right pleura was, therefore, opened. The descending aorta was seen and prepared for anastomosis. Heparinization was performed (100 IU/kg). A Dacron graft, 20 mm in diameter, was anastomosed end-to-side with a side clamp from the ascending aorta. The graft was, then, extended from the right posterolateral side of the pericardium to the descending aorta following an extrapericardial path. The distal part of the graft was anastomosed end-to-side with a side clamp from the descending aorta (Fig. 3). Subsequently, the sternum was closed, and the patient was repositioned. Left posterolateral thoracotomy was performed through the fourth intercostal space. The left arch and the left subclavian artery were looped, and the ligamentum arteriosum was divided. The posterior aortic arch was also divided just distal to the left subclavian artery origin. Both ends of the aortic stump were repaired primarily. Residual scarring was seen between the esophagus and the surrounding tissues. The esophagus was cleared of these adhesions. Esophageal compression was significantly relieved.

The patient was followed up in the intensive care unit for 36 hours. Postoperative CT imaging revealed the disappearance of the posterior pressure on the trachea and the esophagus (Fig 2A). He recovered without any problems and was discharged with acetylsalicylic acid. His respiratory and swallowing problems completely regressed in the follow-up. The patient has been asymptomatic for 2 years at the time of writing this manuscript.

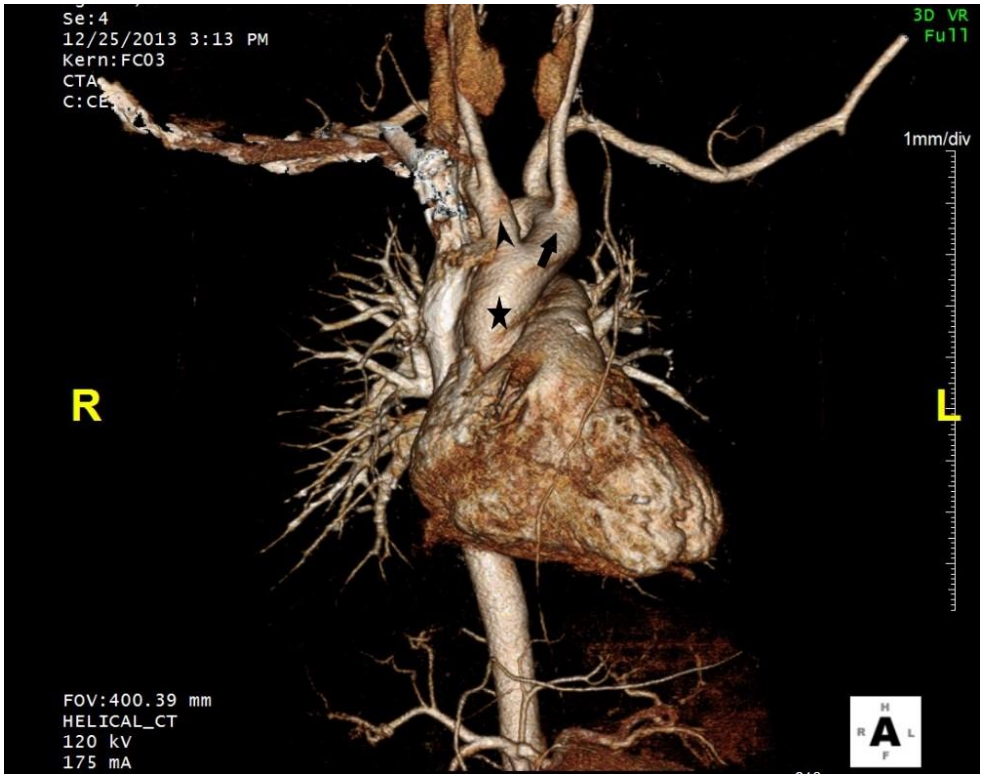

Figure 1: The 3D computed tomography angiography image shows the continuity of the ascending aorta (black mark) as the left-anterior (black arrow) and right-posterior (black arrowhead) arcus. While the right arch gives the right common carotid and right subclavian artery branches separately, the left arch gives the left common carotid artery and left subclavian artery branches.

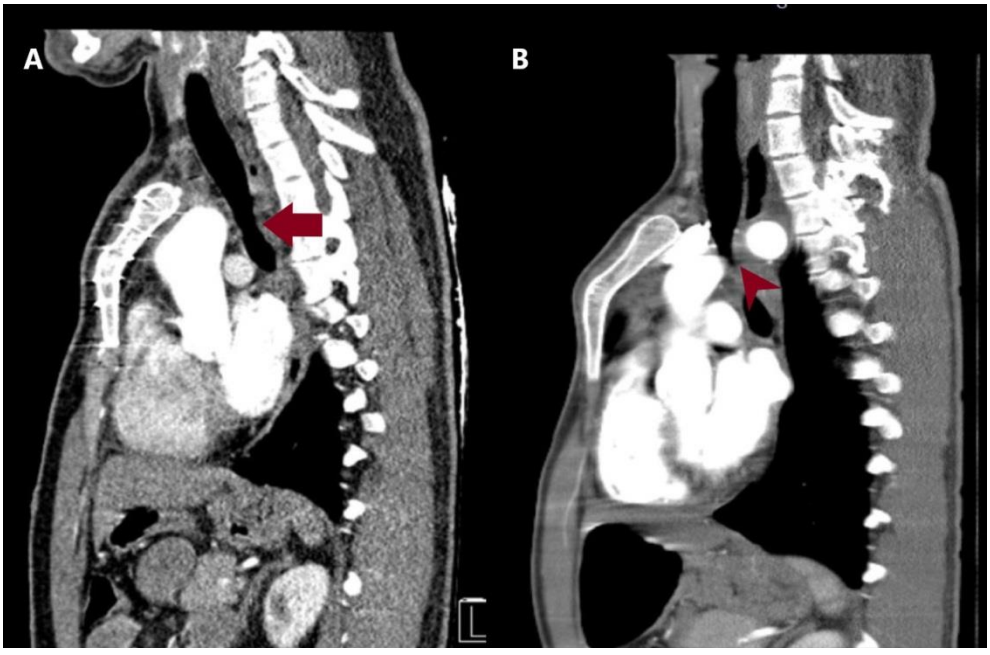

Figure 2: The computed tomography images of the patient after the first operation (B) and after the second operation (B) are presented herein. (A) The tracheal compression has disappeared after extra-anatomic (from ascending to descending aorta) bypass. (B) The tracheal compression continues due to only anterior arch ligation in the first operation.

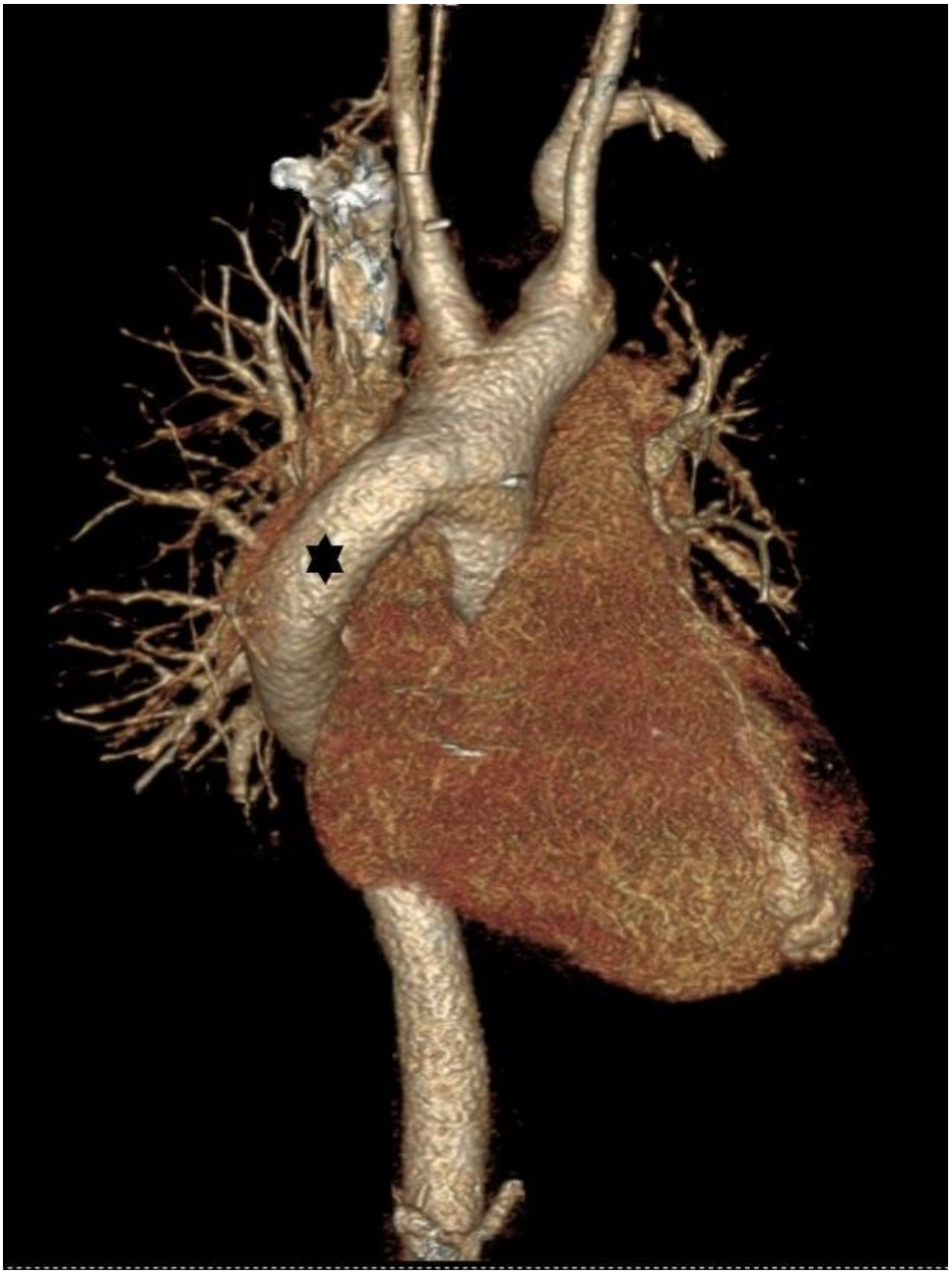

Figure 3: The 3D computed tomography image of the patient after the second operation shows the Dacron graft extension on the right side of the heart from the distal portion of the ascending aorta to the descending aorta (black mark).

DISCUSSION

The vascular ring is a rare anomaly that shows significant improvement with effective surgical treatment. Embryologically, the dorsal aorta is fused and resorbed following a path that forms the usual left aortic arch and the left descending aorta. Still, failure in the resorption phase will result in a series of vascular loops or any of the cases of a pulmonary artery sling. Vascular rings tend to narrow the trachea, the esophagus, or both and can cause significant obstructive respiratory tract or esophageal symptoms.[1] Most patients with vascular rings become symptomatic immediately after birth, and more than half undergo surgery in the first year of their life,

and almost all before the age of 5. Generally, these patients present more respiratory tract-related symptoms due to tracheal compression than swallowing symptoms.[2]

The most common symptoms are stridor, cough, and recurrent respiratory tract infections. Dysphagia is a less common symptom. Patients with vascular rings try to tolerate this situation by chewing food slowly, drinking plenty of fluids during feeding, and feeding very slowly. Our patient had severe swallowing difficulties and a feeling of choking while eating solid foods, as well as complaints of inability to gain weight and dyspnea. He was diagnosed with asthma and reflux many times, for which he received treatment. In the daily practice of pediatricians, vascular rings may be overlooked at the first visit of patients. Nevertheless, the secondary compression of vascular rings should be considered in patients who are unresponsive to medical treatment and have problems with swallowing and dyspnea.

The vascular ring diagnosis is made with barium esophageal radiography and bronchoscopy when access to CT and magnetic resonance imaging is difficult. However, nowadays, CT is often sufficient for a definitive diagnosis. We also confirmed the diagnosis with CT in our patient. It is vital to note that the ligamentum arteriosum can also be viewed with magnetic resonance imaging, especially in redo patients.[3]

Preoperative detailed evaluation of patients diagnosed with the vascular ring is essential. During the decision-making as regards the surgical strategy, the dominant arcus should be identified, and the non-dominant arch should be divided. In the literature, it has been reported that posterior arch division confers better anatomical results if both arches are equal to the aorta.[4] In our patient, both arches were equal, but in the first operation, the anterior (right) arch division did not relieve the pressure on the posterior part of the trachea and the esophagus. Additionally, the direction of the descending aorta is significant. In our patient, the descending aorta was on the right side of the spinal column. The right-sided descending aorta formed a fold with the posterior arch aorta and caused compression. The patient was temporarily well (3 y), and then the symptoms resumed. The reason for this situation may be the re-development of adhesions between the trachea and the esophagus in the long postoperative period.

Patients requiring reoperation after vascular ring surgery have been reported in the literature. The most common situations requiring re-intervention are the Kommerell diverticulum, the circumflex aorta, residual scarring, and tracheobronchomalacia requiring aortopexy.[5] In our patient, we defined the other missing procedures in the first operation as follows: ligamentum arteriosum division was not performed, dissection and release were inadequate, and arteriopexy was not applied to the innominate artery and stump. The patient-related factor was that he was operated on in the late period. We also noticed that the pleura was closed with PROLENE sutures in the first surgery. In vascular ring surgery, the scar tissue that will develop in the suture line causes the compression to continue. Therefore, the pleura should be left open.

In the presence of recurrent symptoms after vascular ring surgery, we applied the extra-anatomic bypass protocol from the ascending aorta to the descending aorta in our patient. This procedure was first described by Vijanayagar[6] in the 1980s. A patient with postductal aortic coarctation was operated on with this technique. Konstantinov et al[7] used this procedure in redo vascular ring surgery for a similar indication. The patient presented by Konstantinov and colleagues had a right

aortic arch and an abnormal left subclavian artery. The third operation on the patient, who had undergone surgery twice before and had recurrent symptoms, was performed with this technique. However, the operation was performed with cardiopulmonary bypass (CPB) and total circulatory arrest. In our patient, we provided access via the right pleural front of the pericardium as the descending aorta descends from the right side of the spinal column. We did not need to position the heart, and we performed the procedure without CPB. We carried out proximal and then distal anastomosis with a side-biting clamp. The proximal anastomosis of the graft should be made as high as possible in the ascending aorta because it should be kept in mind that if the patient needs to undergo another cardiac surgery later, an appropriate area for a cross-clamp will be required.

In conclusion, the need for reintervention is substantial due to recurrent symptoms after vascular ring surgery. Symptoms recur in 5% to 10% of patients, and reintervention is required. In such cases, extra-anatomic bypass may be a good option for the appropriate patient group. With appropriate anatomical conditions and adequate exploration, the procedure can be performed without CPB.

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Conflict of Interest

None declared

Ethical Approval

Not applicable since this study does not contain patient information

Informed Consent

Voluntary informed consent was obtained from the patient.

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