Surgical treatment of double aortic arch

Çift arkus aortun cerrahi tedavisi

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Background: This study aims to describe the management and surgical outcomes of patients with isolated double aortic arch (DAA).

Methods: Data regarding presentation, management, and surgical outcomes of 11 patients (8 girls, 3 boys; median age of 18.6 months) operated between January 2006 and January 2012 due to DAA were evaluated retrospectively. Duration of the reversal of symptoms was recorded throughout the follow-up period.

Results: The median age at the onset of symptoms was 3.5 months (birth to 4 years). Gastrointestinal symptoms were present in all and respiratory symptoms were present in 45.5% of patients, with difficulty in feeding and recurrent respiratory tract infections being the most common symptoms, respectively. All patients underwent corrective surgery successfully. No early or late death occurred. No reoperations were required during follow-ups. Symptoms involving gastrointestinal tract disappeared in all patients (100%) in mean 6.4±4.3 months, whereas respiratory symptoms disappeared in four patients (80%) in 11.4±7.2 months.

Conclusion: Today, results of surgical treatment of DAA are satisfying. Following surgery, while gastrointestinal symptoms disappeared in all patients regardless of age, complete relief of respiratory symptoms requires early intervention.

Keywords: Double aortic arch; oesophageal stenosis; tracheal stenosis; vascular ring.

Amaç: Bu çalışmada, izole çift arkus aort (ÇAA) olgularında tedavi yaklaşımı ve cerrahi sonrası sonuçları tanımlandı

Çalışma planı: Ocak 2006 - Ocak 2012 tarihleri arasında ÇAA nedeniyle ameliyat edilen 11 hastanın (8 kız, 3 erkek; ort. yaş 18.6 ay) başvuru bilgileri, tedavileri ve ameliyat sonuçları ile ilgili veriler retrospektif olarak değerlendirildi. Semptomların düzelme süresi takip dönemi boyunca kaydedildi.

Bulgular: Semptomların başlangıcında ortalama yaş 3.5 aydı (doğumdan 4 yaşa kadar). Gastrointestinal semptomlar tüm hastalarda mevcutken respiratuar semptomlar hastaların %45.5'inde bulunmaktaydı ve en sık görülen semptomlar sırasıyla beslenme güçlüğü ve tekrarlayan solunum yolu enfeksiyonlarıydı. Tüm hastalara düzeltici cerrahi başarıyla uygulandı. Erken veya geç ölüm meydana gelmedi. Takiplerde yeniden cerrahi ihtiyacı olmadı. Gastrointestinal sistemi içeren semptomlar tüm hastalarda (%100) ortalama 6.4±4.3 ayda düzelirken respiratuar semptomlar dört hastada (%80) ortalama 11.4±7.2 ayda düzeldi.

Sonuç: Günümüzde, ÇAA'nın cerrahi tedavisinin sonuçları tatmin edicidir. Cerrahi sonrasında, yaştan bağımsız olarak tüm hastalarda gastrointestinal semptomlar kaybolurken respiratuar semptomların tam düzelmesi için erken girişim yapılması gerekmektedir.

Anahtar sözcükler: Çift aort arkı; özofagus darlığı; trakea darlığı; vasküler halka.



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Double aortic arch (DAA), first described by Hommel in 1737.[1] is a relatively rare congenital cardiovascular malformation characterized by the formation of two aortic arches caused by the persistence of the right eighth segment of the dorsal aortic root during the development of the branchial arteries.[2] In DAA, the two aortic arches form an anatomically complete vascular ring around the trachea and/or esophagus,[3] and in more than 75% of cases, there is a larger (dominant) right arch in back of and a smaller (hypoplastic) left aortic arch in front of the trachea/ esophagus.[4] The two arches then join to form the descending aorta, which is usually on the left side but may also be right-sided or at the midline. [5] In some patients, the end of the smaller left aortic arch closes (left aortic arch), and the vascular tissue becomes a fibrous cord. [6-9] In addition, the vascular ring can compress the trachea and/or esophagus. Although this is a vascular malformation, patients may also suffer from respiratory and gastrointestinal symptoms, including dyspnea, stridor, cough, recurrent respiratory tract infections, or difficulty in feeding in infancy and childhood as well.[10,11] Double aortic arch is the most common type of congenital vascular ring anomaly^[6,7] with a wide prevalence rate of between 18 and 77% and a calculated weighted mean of 48% among all vascular rings.[12] Diagnosis can often be suspected via chest X-ray, barium esophagography, or echocardiography, but computed tomography (CT) or magnetic resonance imaging (MRI) shows the relationship of the aortic arches to the trachea and esophagus and the degree of tracheal narrowing. In addition, a bronchoscopy can be useful for internally assessing the degree of tracheomalacia. Surgical treatment is indicated in all symptomatic patients, but the risk of mortality or significant morbidity after the surgical division of the lesser arch is low in the current era. However, the preoperative degree of tracheomalacia can dramatically impact postoperative recovery. Moreover, in certain patients, it may take from several months to as much as two years for the obstructive respiratory symptoms (e.g., wheezing) to disappear.

PATIENTS AND METHODS

In the present study, we sought to review the surgical management and postoperative outcomes of patients who underwent corrective surgery due to isolated DAA. Hence, we retrospectively reviewed the medical records of 11 patients (8 girls, 3 boys; median age of 18.6 months) with this malformation who were operated on between January 2006 and January 2012 at the Dr. Siyami Ersek Thoracic and Cardiovacular Surgery Training and Research Hospital. This study protocol was approved by the hospital ethics committee in 2012.

Table 1 shows the patients' age at the first onset of symptoms, gender, which diagnostic tools were used, and which structures were compressed by the vascular ring, and the patient's age at the time of surgery, length of intensive care unit (ICU) and hospital stays, and presenting symptoms are given in Table 2.

The median age at the first onset of symptoms was three and a half months (birth-4 years old), and the median age at surgery was 18.6 months (range 3 months-4 years old) (Table 1 and 2).

All of the patients, except for the two who were three and five months old, were referred to our clinic

Table 1. Characteristics of patients who underwent corrective surgery for double aortic arch

Patient	Age at first onset of symptoms (months)	Gender	Diagnostic tools	Structures compressed by the vascular ring		
1	1.5	F	Echocardiography + CT + barium esophagography	Trachea + esophagus		
2	3	F	Echocardiography + MRI	Esophagus		
3	4.5	F	Echocardiography	Esophagus		
4	3.5	M	Echocardiography + CT + barium esophagography	Esophagus		
5	2.5	F	Echocardiography + CT	Trachea + esophagus		
6	1	F	Echocardiography + CT	Trachea + esophagus		
7	5.5	F	Echocardiography + MRI	Esophagus		
8	2	M	Echocardiography + CT	Trachea + esophagus		
9	8	F	Echocardiography + MRI + barium esophagography	Esophagus		
10	1	M	Echocardiography + CT	Trachea + esophagus		
11	6.5	F	Echocardiography	Esophagus		

F: Female, M: Male; CT: Computed tomography; MRI: Magnetic resonance imaging.

Table 2. Follow-up of patients who underwent corrective surgery for double aortic arch

Patient	Age at surgery (months)	ICU stay (days)	Hospital stay (days)	Dyspnea	Stridor	Cough	Recurrent respiratory tract infections	Difficulty in feeding	Weight loss
1	34	1	9	X	Х	Х	20	11	3
2	8	1	4	X	X	X	X	3	2
3	25	1	7	X	X	X	X	9	3
4	35	1	6	X	X	X	X	12	4
5	16	1	6	9	X	X	X	4	2
6	4	2	7	11	X	X	X	1	1
7	16	2	4	X	X	X	X	6	3
8	3	2	8	X	1	X	X	1	2
9	48	1	5	X	X	X	X	13	3
10	5	2	9	X	X	16	X	4	1
11	11	1	7	X	X	X	X	6	2

ICU: Intensive care unit; x: Not present preoperatively

from other hospitals that did not normally perform pediatric cardiac surgery.

Several diagnostic modalities, including X-ray roentgenograms, barium esophagography, bronchoscopies, and bronchography, had been previously performed because of the initial suspicion of DAA in most of the patients. While we used echocardiography at first to check for the presence of this disease, CT and MRI were used to verify the diagnosis (Figure 1). In addition, three patients had also previously undergone barium esophagography (Table 1) before being referred to our hospital to reveal the cause of difficulty in feeding. Using the aforementioned modalities, DAA was diagnosed in all of our patients with a right-sided (posterior) dominant and a smaller left-sided (anterior) nondominant aortic arch.

Gastrointestinal symptoms, (e.g., difficulty in feeding) were identified in all of the patients, and respiratory symptoms, including dyspnea, stridor, cough, and recurrent respiratory tract infections, were also observed in five of the study participants preoperatively. In addition, common symptoms like difficulty in swallowing following the start of solid foods were noted in the infants, and increased respiratory distress in the extension position was also encountered in half of the patients.

Although there was no history of premature delivery, low birth weight or other extracardiac abnormalities present in the patients, weakness and difficulty in gaining weight were common complaints. Furthermore, they were all below the normal ranges of their peers in both weight and height.

Compression on the esophagus was only seen in six patients (54.5%) and on both the esophagus and airway in five others (45.5%) (Table 1), with the median age

of those with respiratory symptoms being five months old (range 3-34 months). This was significantly lower than the group as a whole. One of the patients with respiratory symptoms had been on ventilator support for two weeks before presentation, and every attempt for extubation had resulted in instant respiratory distress. Although apnea and cyanosis are among the most important symptoms that affect mortality or morbidity in this pathology, these symptoms were not observed in our patients.

Our patients' preoperative anesthetic regimen was similar to all pediatric cardiac operations and included general anesthesia with endotracheal intubation,



Figure 1. Angiography via magnetic resonance imaging showing the anterior (A) and posterior (P) aortic arches.

invasive arterial and venous catheterization lines for hemodynamic monitorization, brain monitorization using near-infrared spectroscopy (NIRS), and standard sedative agents. In addition, arterial monitorization was done through the right radial artery because of the dominant right aortic arch observed in all of the patients. The surgery was initiated via a left posterolateral thoracotomy performed on the fourth intercostal space on the side of the descending aorta in all of the cases (Figure 2). Selective ventilation of the lungs was not needed since the left lung was easily removed from the surgical area using a lung retractor. The arterial pulses of both arms and one leg were also monitored using pulse oximetry before the occlusion of the lesser aortic arch, and an occlusion test was carried out on each patient to ensure the identification of the lesser aortic arch prior to division.

The patent ductus arteriosus (PDA) distal to the left subclavian artery was double ligated and divided. In addition, the left (anterior, nondominant) arch was hypoplastic in all of the cases, and the anterior arch was ligated at a distance on the left side of the esophagus and at the junction with the descending aorta (right side of the esophagus) using silk ligatures. Dividing the anterior arch between these ligatures broke down the vascular ring formed by the right and left arches (Figure 3). Due to the hypoplastic, soft nature of the anterior arch, ligation of this rudimentary vessel was enough for a secure closure before division, and no oversewn sutures were needed.

The ligation or division of the PDA and anterior arch did not result in any hemodynamic alteration



Figure 2. Surgical view via a left thoracotomy showing the anterior (A) and posterior (P) aortic arches, descending aorta (D), patent ductus arteriosus (x) and left subclavian artery (S).

because of the small amount of blood flow through the nondominant anterior arch. Furthermore, aortic crossclamping and cardiac arrest were not necessary since the dominant posterior aortic arch maintained the descending aortic blood flow throughout the procedure. Moreover, no patient required heparinization because the procedure only consisted of the division of a vascular structure, so there was not need for vascular anastomosis or cardiopulmonary bypass (CPB). We freed the esophagus underneath the divided left aortic arch from the adjacent adherent tissues as much as possible (Figure 4), and we took extreme care during the procedure to avoid any damage to the neighboring structures (i.e., the vagus, phrenic, and recurrent laryngeal nerves, esophagus, and any lymphatic or vascular structures. We also did not perform any intraoperative intercostal neural blockage on the patients because the postoperative intravenous analgesic therapy was satisfactory.

Postoperative intensive care included standard follow-up involved mechanical ventilation, invasive arterial and noninvasive electrocardiography, a thoracic roentgenogram, and the monitoring of urinary output and temperature. Following extubation, 1 mg/kg of daily oral captopril was begun for all of the patients.

In the early follow-up period, the ICU and hospital stay durations were recorded along with the elapsed

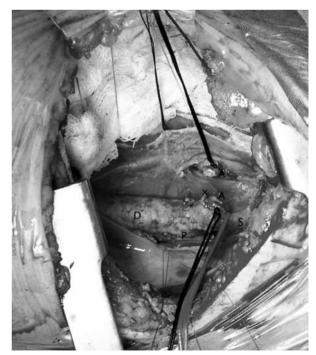


Figure 3. Division of the anterior (A) arch and patent ductus arteriosus (x) distal to the left subclavian artery (S).

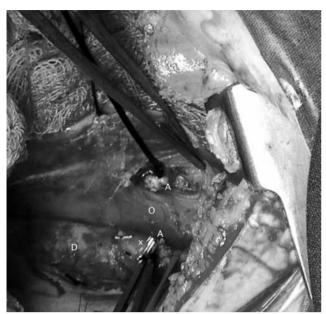


Figure 4. The esophagus (o) underneath the divided anterior arch was freed from the adjacent adherent tissues. A: Anterior arch; D: Descending aorta; x: Patent ductus arteriosus.

time until the patients' symptoms disappeared (Table 2). Long-term follow-up was based on check-ups performed every three months, and clinical investigation and echocardiography were also performed.

All statistical analyses were conducted using the PASW Statistics version 18.0 for Windows software program (SPSS Inc., Chicago, IL, USA). The data was given as frequencies, medians with ranges, and means with standard deviations (SDs), as appropriate, and the factors associated with mortality were compared using Fisher's exact test for a small population. A p<0.05 was considered to be statistically significant.

RESULTS

All of the patients underwent corrective surgery lasting between 1.5 and 3.1 hours, and they had an uneventful recovery. No patient suffered from prolonged chest tube drainage, and there were no intraoperative and postoperative complications except for one patient who developed Horner's syndrome in the early postoperative period. This child was treated successfully with 1 mg/kg methylprednisolone administered for five days. Furthermore, there were no early or late deaths, and none required a reoperation in the early or late follow-up periods.

The mean duration of the postoperative mechanical ventilation was 5.4 ± 3.2 hours, and the mean times of the ICU and hospital stays were 1.4 ± 0.5 and

 6.5 ± 1.7 days, respectively. In addition, there was no significant difference between the patients who had only gastrointestinal tract symptoms and those with both gastrointestinal tract and respiratory symptoms when the postoperative mechanical ventilation and ICU stay durations were evaluated (p>0.1). However, the mean length of hospital stay was significantly higher (7.8 \pm 1.2 days) in the patients with both gastrointestinal tract and respiratory symptoms than those with only gastrointestinal tract symptoms (5.5 \pm 1.5 days) (p<0.05).

The patients had a median age of 5.1 years (2.2-10.5 years), and their average follow-up time was 2.7±4.3 years. All experienced relief of their gastrointestinal tract symptoms at a mean of 6.4±4.3 months (range 1-13 months) (Table 2). Moreover, although the common complaints of weakness and difficulty in gaining weight had disappeared, the patients continued to be below the normal ranges of their peers in both weight and height at the last follow-up.

The respiratory symptoms were relieved at a mean of 11.4 ± 7.2 months (range 1-20 months) in four patients, but one patient was still suffering from recurrent bronchitis at the last follow-up, which was significantly longer than the time it took to relieve the gastrointestinal symptoms (p<0.05) (Table 2).

DISCUSSION

The phrase "vascular ring" has been used to refer to a collection of congenital vascular variations that encircle and compress the trachea and/or esophagus. Patients with compression of these structures present with stridor and/or difficulty in feeding in the neonatal period or even later in early childhood depending on the degree of compression. Two classic vascular rings are formed by the right aortic arch with the left ligamentum arteriosum and a DAA,[3] with the latter being the most common clinically recognized form. [6,7] Anterior to the trachea, the ascending aorta divides into left and right arches that pass on either side of the trachea. In functional DAAs, the left common carotid and subclavian arteries arise from the left arch, and the right common carotid and subclavian arteries arise from the right arch. In more than 75% of cases, the recurrent laryngeal nerves pass under both of these arches.[8,9] The descending aorta may be situated at the mid-line, or to the left.^[5] The two arches can be of equal size, or the right (posterior) arch might be the larger main aortic arch with the left (anterior or pretracheal) arch being comparatively smaller.[13] As described in previous studies, both arches are usually

patent, and in patients with one aortic arch larger than the other, the right arch is dominant in 70-75% of all cases.^[4,14,15]

Surgical repair is usually achieved through a left thoracotomy in patients with a dominant right aortic arch, the most common form of DAA,^[11,16-18] and this is usually performed via the fourth intercostal space.^[19] Prior to the division, the esophagus and trachea should be freed from the adjacent structures. The arterial pulses of both arms and one leg should also be monitored before the occlusion, and an occlusion test must be performed to verify the existence of the lesser aortic arch prior to the division.^[20]

Α right posterolateral thoracotomy is recommended for patients with a left aortic arch, right-sided descending thoracic aorta, and right ductus or ligamentum arteriosum. This procedure is also preferred when DAA with atresia of the right posterior segment is present or when anastomosis of an aberrant right subclavian artery to the ascending aorta is performed.[21] Additionally, in patients with innominate artery compression, this approach is favored from the right side in conjunction with the suspension of the innominate artery to the sternum.[22] However, a median sternotomy is recommended when concomitant repair of intracardiac defects is performed.[21,23]

In this study, all of patients had DAAs with a dominant right-sided (posterior) and a smaller left (anterior) nondominant aortic arch. The nondominant anterior arch was rudimentary, and dividing this rudimentary arch via a left thoracotomy relieved the compression on the esophagus and trachea. We did not encounter any patient with completely equal right and left aortic arches. Due to the rudimentary and elastic nature of the anterior arch, ligation of this structure was sufficient for closure before the division. In patients with equal right and left aortic arches, the anterior arch may be well developed and almost normal in size, and oversewing the suture closure may be required after cross-clamping the anterior arch division in this area.

Double aortic arches reportedly manifest earlier than other varieties of vascular rings and have symptoms of stridor, dyspnea, cough, and recurrent respiratory infections. [21,24,25] In this study the long interval between the first onset of symptoms and the surgery was due to the delay in establishing the correct diagnosis. This was primarily caused by the ambiguous nature of the symptoms associated with DAA, which commonly leads physicians to diagnose

this phenomenon as a respiratory tract infection or metabolic disease that is responsible for the patient's inability to gain weight. Similar to our series, other studies have detected respiratory symptoms in only in a small number of patients at birth or in the first years of life. However, gastrointestinal symptoms, such as difficulty in swallowing, vomiting, and feeding intolerance related to esophageal compression, have been identified as the most common symptoms leading to a correct diagnosis of DAA.^[5,8,11,21,24] To prevent any delay in diagnosis, all patients with suspicious symptoms must be evaluated thoroughly with advanced diagnostic modalities. If sufficient techniques are not available, the patients should then be referred to specialized centers.

Postoperative complications following DAA repair include bleeding, vocal cord paralysis, pneumonia, pneumothorax, chylothorax, difficulty in feeding, and residual respiratory obstruction. In fact, residual respiratory complaints have been noted in up to 54% of patients with this malformation. [11,17,18] If there is significant long-term tracheal compression, there may be damage to the tracheal rings as well as a localized area of tracheomalacia. [26]

In our series, the only postoperative complication was Horner's syndrome in one patient in the early postoperative period. However, the postoperative length of hospital stay was significantly longer for those having respiratory symptoms (p<0.05), and the duration of relief for the patients with respiratory symptoms was significantly longer than for those with symptoms involving the gastrointestinal tract (p<0.05) (Table 2). These findings served as a reminder of the possible damage in the tracheobronchial system for our patients who had suffered for a relatively lengthy time with respiratory symptoms.

Conventional chest X-rays may show the indentation of the tracheal shadow, retrotracheal opacity, and anterior tracheal bowing, and specific radiological signs have been described for patients who underwent barium esophagography.^[27] These include bilateral persistent extrinsic compressions of the esophagus in the anteroposterior (AP) view in which the dominant arch causes a deeper superior indentation and a deep posterior indentation in the lateral and oblique views. A barium swallow is diagnostic in the majority of these cases, but CT and MRIs are being increasingly used in the diagnosis and evaluation of DAA[16] because they can show the four-vessel sign in the superior mediastinum due to the presence of separate subclavian and common carotid arteries on both sides. Presently, angiography is rarely indicated or necessary to adequately evaluate aortic arch anomalies, but echocardiography is recommended to rule out associated congenital cardiac defects. [11,16,18,23] Thus, although X-ray roentgenograms, barium esophagography, bronchoscopies or bronchography are helpful when DAA is suspected, the definitive diagnosis is made by CT or MRI and then supplemented by echocardiography. However, simple diagnostic modalities like X-ray roentgenograms may still be the only method available in rural districts, and these can lead to further investigations to establish the correct diagnosis.

Surgical division of the vascular ring is indicated for any patient that presents with symptoms or has tracheal and/or esophageal compression. There are no other treatment modalities other than surgical intervention to divide the ring.^[28-30] The goal is to relieve this compression by dividing the smaller of the two arches at a site that does not compromise the blood flow to the head vessels.^[5] Although division of the lesser aortic arch is adequate in patients with a dominant right or left aortic arch, division of the right aortic arch may be preferred in patients with equal right and left aortic arches to achieve a more natural anatomy.

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