Pseudoaneurysm Complicating Anterior Arch of Congenital Double Aortic Arch

Atefeh Asadmobini[1], Feridoun Sabzi[2], Aghigh Heidari[3]

[1] *Department of Cardiology, Imam Ali Hospital, Kermanshah University of Medical Sciences, Kermanshah, Iran*

[2] *Department of Cardiovascular Surgery, Imam Ali Hospital, Kermanshah University of Medical Sciences, Kermanshah, Iran*

[3] *Department of Anesthesiology, Imam Ali Hospital, Kermanshah University of Medical Sciences, Kermanshah, Iran*

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Abstract- Aortic rupture and pseudoaneurysm formation in the anterior arch of congenital double aortic arch anomaly associated with a coarctation is an exceedingly rare complication of aortic vascular ring. This is a case report of a double aortic arch with an anterior arch pseudoaneurysm, presenting with chest pain, respiratory distress, and hypotension. The patient underwent open-heart surgery with resection of the anterior arch pseudoaneurysm and bypassing of the coarctation with ascending aorta to descending aorta connection with a Dacron tube graft. Posterior aortic arch entering to pseudoaneurysm by a vertical artery also connected to the main Dacron tube by another small size of the Dacron tube. The postoperative course was complicated by chylothorax and respiratory distress that managed with prolonged preservation of chest tubes, chest physiotherapy, and low lipid content diet. One year follow-up with C_T angiography revealed the patency of both the Dacron tube and patient was in good condition.

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Keywords: Congenital heart disease; Double arc; Aneurism

Introduction

Most patients with double aortic arch anomaly presenting with tracheoesophageal compression in early life and vascular complication is a very rare event (1). Aneurysm formation in the aortic arch also may be associated with thromboemboli, rupture, and pseudoaneurysm formation (2). Association of the double arch with coarctation and ruptured aneurysm of the anterior arch is an exceptionally rare phenomenon. In our case, a ruptured aneurysm of anterior arch disturbed normal anatomy of the double arch, and other mediastinal organ and transesophageal echocardiography (TEE) showed only a ruptured descending aortic pesudo aneurysm with coarctation. The proper diagnosis was made when the left thoracotomy exploration revealed absence of branching of the arch vessels from aneurysmal anterior arch vessel.

Case Report

A 26-year-old bodybuilder male was referred to our institution with hypotension, reduced conciseness, chest pain, discoloration of left foot, fingers, frequent episode of fainting, for evaluation, and management of ruptured thoracic aneurysm. A Physical exam revealed a pulse of 120 beats/min and blood pressure of 80/40 mmHg. The patient was hypotensive, tachypneic, and had diffuse left lung crepitating rales. The ECG revealed sinus rhythm with hypertrophy of both ventricles. The Results of the initial tests of cardiac enzymes were normal. A chest x-ray showed a large mass in the left hemithorax. Emergency TEE, showed normal cardiac anatomy and ascending aorta, but revealed a huge pseudoaneurysm in the aortic isthmus in the precoarctation segment of descending aorta filled with thrombus and with normal arch vessels. The posterior aortic arch with its related branches was not detected in TEE. The distal trachea and proximal left bronchus were not compressed by a ruptured anterior arch pseudoaneurysm. The posterior arch ended to the innominate artery and, after debranching of the left carotid artery and subclavian artery, bends downward and connect to descending aorta in the pre coarctation portion of the descending aorta. The patient underwent emergency aneurysm resection through a left posterolateral thoracotomy approach, although the right femoral artery and vein prepared for possible cannulation (Figure 1). The posterior aortic arch, giving rise to the innominate artery, left subclavian artery, and left common carotid artery (Figure 2). The

Corresponding Author: A. Heidari
Department of Anesthesiology, Imam Ali Hospital, Kermanshah University of Medical Sciences, Kermanshah, Iran
Tel: +98 918333733, Fax: +98 8313836004, E-mail address aghigh heidari@yahoo.com

left aortic arch was located anteriorly, giving rise to the large pseudoaneurysm that connected to descending aorta in pre coarctation area (Figure 3). The decision was made to prevent further possible distal thromboemboli of the thrombotic pseudoaneurysm by distal control (descending aorta ligation) and bypass of ascending aorta to the distal descending aorta. However, distal control of descending aorta below the aneurysm easily performed, but severe inflammatory reaction and hematoma of the perineurial sac obviate access to post subclavian part of descending thoracic aorta for proximal anastomosis (Figure 4).

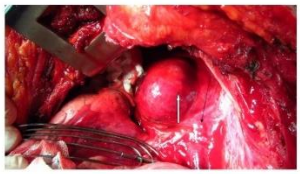

Figure 1. Shows large pseudoaneurysm that distorts aortic arch and descending aorta, the posterior arch was also hidden below the aneurysm as coarctation site. (left anterolateral thoracotomy)

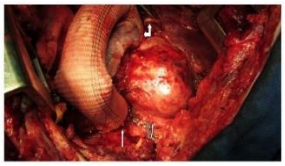

Figure 2. Revealed exclusion of aneurysm (curved arrow) by Dacron tube between ascending aorta (two-headed white arrow) and sub coarctation descending aorta (vertical with arrow)

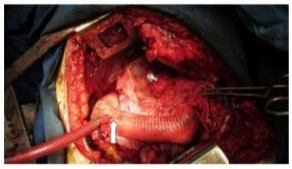

Figure 3. Shows cannulation of bypassed Dacron tube (with arrow) for arterial line combined with right femoral vein to constitute cardiopulmonary bypass in left thoracotomy view

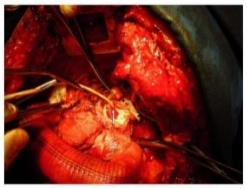

Figure 4. Opening of the pseudoaneurysm (black arrows) after the constitution of CPB. Brisk bleeding from the small mouth of the anterior arch vessel connected to posterior arch temporary controlled by folly catheter and then close with 4/0 PROLENE® suture (vertical white arrows)

After distal anastomosis of Dacron tube (20 mm) to distal of coarctation in the normal descending aorta, thoracotomy incision was approximated by tower clips, and the patient returned to the supine position. A median sternotomy was performed and proximal of Dacron graft connected to the left lateral side of ascending aorta. Dissection of the arch vessels revealed that these vessels, i.e., innominate, left common carotid, and left subclavian artery, to arise from the posterior arch that with vertical artery connected to the aneurismal anterior arch. The left subclavian artery arises from this vertical artery. A median sternotomy was closed, and the patient repositioned again in lateral thoracotomy. Following right femoral vein cannulation, and interposition Dacron graft cannulation, CPB constituted, and the patient further cooled to 20 centigrade, with total cardiopulmonary arrest. The wall of pseudoaneurysm was opened, and ball shape clot and inflamed tissues were removed. The mouth of the aneurismal anterior aortic arch in connection site to the posterior arch was oversewn by 4/0 proline (Figure 5). The vertical artery was ligated in the entrance to aneurysmal sac and further anastomosed to main Dacron graft by small Dacron tube (number 6 mm) graft. The postoperative course was complicated by chylothorax that begins in 2nd days of operation. The dependency on mechanical ventilation has lasted for three days that during this course, respiratory tract managed by adding a peep to the respiratory parameter and the observance of good hygiene the chylothorax was resolved in the 8th days of operation with limitation of fat intake and preserving of the chest tube. Foot discoloration may be caused by multi factors such as micro thromboemboli, hypotension, or coarctation that successfully treated by bypassing coarctation and concomitant anticoagulation therapy.

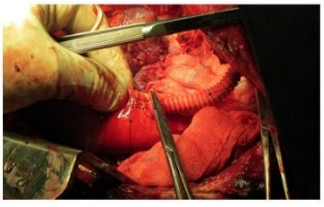

Figure 5. Shows anastomosis of the posterior arch to the main Dacron tube (vertical black arrow) by small Dacron tube (transverse black arrow) to establish continuity of posterior arch vessels to the distal descending aorta

Discussion

The most common complication of the double aortic

arch is compressing the trachea and esophagus that are encircled by these arches and their derivative branches. Embryologically, the ventral and dorsal aorta are connected by aortic arches, which may persist as a double arch anomaly or the anterior arch involutes to perform normal aortic arch and its branches. Other complication of these anomaly includes hypoplasia of arch branches, dissection, and aneurysm. But rupture and pseudoaneurysm of the anterior arch were not reported in the medical literature (3). Ito reported a patient with double aortic arch and left atretic arch proximal to the left common carotid artery (subtype 4), and also tapering and aneurysm of the left arch distal to the left subclavian artery. To the authors' knowledge, this case is the first report of the double arch anomaly with left or anterior arch atresia. Satoshi Ienaga showed, presence of an aneurysm that involving ascending aorta and the left (anterior) arch of the double aortic arch. Surgical repair was performed by removing the ascending aneurysmal aorta and left aortic arch, and a Dacron tube was inserted between the ascending aorta and posterior aortic arch (4). Tomoki Choh reported a type 4 Stewart double arch anomaly that defined as the presence of ascending thoracic aortic aneurysm with incomplete double aortic arch associated with PDA. Resection of the anterior arch aneurysm with Dacron replacement of the ascending aorta was undertaking under CPB institution (5). Liang Y reports a 21-year-old man who was presented with a double aortic arch anomaly associated with ascending aortic aneurysm and aortic valve regurgitation. The patient underwent aortic root replacement and construction of side branches with artificial Dacron tubes (6,7). A review of the literature shows that combination of double aortic arch anomaly with pseudoaneurysm formation has not been reported so far.

Anabolic steroid abusing in bodybuilders is associated with hypertension, and medial degeneration in the great vessels wall. In these cases, weight lifting causes rapid hemodynamic changes that, with a sudden increase in systemic arterial blood pressure in the presence of concomitant aortic medial degeneration may lead to aneurysm formation and rupture.

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