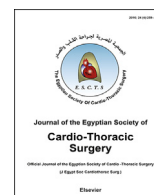


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Vascular rings and slings: A challenging diagnostic and therapeutic rare disease entity



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ABSTRACT

Background: Vascular rings (VRs) are classified as true rings or partial rings. The complete VRs include double aortic arch (DAA) and right aortic arch (RAA) with aberrant left subclavian artery and left ligamentum. The incomplete (VRs) include innominate artery compression and pulmonary artery sling.

Methods: We retrospectively analyzed the data from 90 patients with VRs and pulmonary artery slings diagnosed at Mansoura University Children's Hospital between 2004 to 2016. The initial diagnostic clue might be a right sided aortic arch on a plain x-ray chest. Lateral views were evaluated for retro tracheal opacity, anterior tracheal bowing, and posterior indentation. In our series, bronchoscopy was used for patients with respiratory tract symptoms. Bronchoscopy was performed in 21 patients (23.3%), with finding of external tracheal wall pulsatile compression. Echocardiography was used initially in most cases as the first diagnostic tool followed by Multislice CT scan (MSCT).

Results: The age of operation was significantly earlier in patients with complete VRs versus incomplete vascular rings. By far the commonest variant was the DAA which constituted in our series 57.8%. The second common type was RAA with aberrant left subclavian and left ligamentum which constituted 30%. The third most common type was pulmonary artery sling constituting 6.7%. In our series, Echocardiography was done in 78 cases (85.6%). It was useful not only in initially diagnosing VR anomaly but also it detected associated cardiac anomalies in 4 cases. MSCT was an excellent noninvasive diagnostic modality that was used in 75 cases (83.3%). We operated 90 cases; 79 via left thoracotomy, 10 cases via median sternotomy and one case via right thoracotomy. Among the 90 cases that were operated, there was a single mortality, no cases were complicated by recurrent laryngeal nerve injury and five cases had postoperative chylothorax.

Conclusions: In our series, Echocardiography and MSCT were essential for proper diagnosis and planning for surgery. Left thoracotomy or median sternotomy provided good exposure in the operated cases. Surgery was done with low mortality and morbidity.

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1. Introduction

Vascular rings (VRs) and pulmonary artery slings are a group of anomalies caused by abnormal embryologic development of the branchial arch system. They are characterized by an anomalous branching pattern of the vessels originating from the aortic arches, abnormal positioning of the aortic arch itself, interrupted or supernumerary arches, or anomalous origin of the pulmonary artery from the contra lateral pulmonary artery or ascending aorta [1].

These anomalies cause compression of the trachea, esophagus, or both. VRs may be either asymptomatic or more frequently may present with respiratory and/or gastrointestinal symptoms that depend on the degree of tightness of the ring and on subsequent tracheoesophageal compression. Most children with VRs present with symptoms in the first few months of life and require surgery within the first year of life [2].

They were classified by the society of thoracic surgeons International Congenital Heart Surgery Nomenclature and Database Committee into 2 main types [3]:

1. Complete Rings:

- Double aortic arch: Dominant right - Dominant left - Balanced
- Right arch/left ligamentum: Retro esophageal left subclavian - Mirror image branching

2. Incomplete ring:

- Pulmonary artery sling
- Innominate artery compression syndrome
- Left arch/aberrant right subclavian artery

1.1. Diagnostic studies

There are diagnostic modalities available for evaluation of the child suspected of having a VR. Barium esophagram used to be the only important tool for diagnosing a VR. Bronchoscopy is important diagnostic tool and in the presence of a vascular ring, shows an extrinsic, often pulsatile, compression of the trachea. Echocardiography is useful for making the diagnosis of most of VRs and pulmonary artery slings. Computed tomography (CT) and magnetic resonance imaging (MRI) identify both the vascular structures and the tracheo-bronchial anatomy. Angiography is rarely needed for the diagnosis of VRs.

1.2. Surgical treatment

In general, VRs are approached through a left lateral thoracotomy and do not require cardiopulmonary bypass. In a double aortic arch the smaller of the two arches is divided. Arch division should always be done between vascular clamps with over sewing of the divided stumps, because simple ligation and division has been associated with ligature slippage and subsequent catastrophic hemorrhage [8]. In patients with a right aortic arch (RAA) and an aberrant left subclavian artery the ligamentum arteriosum requires division [9].

The approach to patients with innominate artery compression involves an aortopexy via a median sternotomy incision, affixing the proximal innominate artery to the posterior aspect of the sternum. Another option involves division and re-implantation of the innominate artery [8].

Patients with a pulmonary artery sling are repaired via a median sternotomy approach and with or without the use of cardiopulmonary bypass. The left pulmonary artery is transected at its origin from the right pulmonary artery and re-implanted on the left lateral aspect of the main pulmonary [10].

2. Patients and methods

We retrospectively analyzed the data from 90 patients with vascular rings and pulmonary artery slings diagnosed and operated at Mansoura university children's hospital between 2004 and 2016.

Age, gender, presenting symptoms, imaging modalities, surgical treatment and post-operative course were studied.

2.1. Operative technique

All patients were operated upon by the same surgeon, anesthesia, and nursing teams using the same surgical technique. After anesthetizing the patient; it was our routine technique to have two arterial lines, one in the right arm and one in the lower limb. Also, we used three pulse oxymetries, one on each of the upper limbs, or ear lobes, and one on the toe. This was important in helping determining the dominant arch, especially in cases with balanced arch type. For cases of DAA, the approach was through a posterolateral thoracotomy with sparing of the serratus anterior muscle.

The left lung was retracted anteriorly and downward, the mediastinal pleura was incised using low grade diathermy. The ligamentum arteriosum, left arch, left subclavian and left common carotid arteries were clearly identified and dissected freely

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