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OUTCOMES OF VARIOUS PATIENT PERSONALISED APPROACHES IN VASCULAR RING REPAIR SURGERY-OUR SINGLE CENTRE EXPERIENCE

Cardiothoracis

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ABSTRACT

Background: Vascular rings are part of a larger group of vascular-related compression syndromes. The approach to the diagnosis and management of these syndromes needs to be individualized. This single-institution study assessed the midterm outcomes of patients undergoing vascular ring repair using personalised approach in terms of surgical approach(Midline Sternotomy versus Thoracotomy) and surgical technique(complete division of vascular ring in symptomatic patients to just division of ligamentum arteriosum in asymptomatic patients with vascular ring as a incidental finding). **Methods**: The study included all patients with vascular ring and sling who underwent surgery from 2017 to 2023 at our institution. Patients who underwent concomitant intracardiac repair were also included. Data analysis included demographics, type of anomaly, other congenital heart disease, clinical symptomatology, operative technique and perioperative outcomes. **Results:** Repair through open thoracotomy was performed in 3 patients (27.3%), and median sternotomy in 8(72.7%) patients. Median age at time of repair was 13.9 months (range 3-39 months), and median weight was 5.8 kg (range, 3-12 kg). At the time of operation, 81.8% (9 out of 11) of patients were younger than 1 year and 18.2% (2 out of 11) were older than 3 years. Nine patients (81.8%) were boys. DAA was present in 3, RAA with ALSCA and LLA in 5, and LAA with ARSA in 1 and LPA sling in 2 patients. **Conclusions:** Freedom from reoperation after vascular ring repair was 100% at 5 years . 3 patients who only underwent resection of ligamentum arteriosum and no diverticulum resection or aberrant left subclavian artery reimplantation at the time of surgery were also free from reintervention till the completion of study. Single midline Sternotomy incision provide as good exposure as conventional double incision approach and better post operative outcome in terms of decreased morbidity than the double incision approach.

KEYWORDS

INTRODUCTION :

The term "vascular ring" refers to congenital aortic arch anomalies that lead to compression of the trachea, the esophagus, or both. This compression leads various symptoms such as respiratory distress or swallowing difficulties .

The first description of a vascular ring was given by` anatomist Hommell, who described the anatomy of a double aortic arch in 1737.[1,2] The term "vascular ring" is often attributed to Robert E. Gross who described the autopsy finding in a five-month-old baby in 1931, nearly fifteen years prior to his report on the surgical correction.[3] .The nomenclature and classification systems for vascular rings have varied over time. Vascular rings were classified into 7 types, A through G in a report of Mayo Clinic in 1971 .[4] Later on, in 2000, a simplified grouping system was proposed by Backer and Mavroudis [5] that divides vascular rings into four main categories : double aortic arch, right arch with left ligamentum, innominate artery compression, and pulmonary artery sling. A study done by Mitchell and colleagues ,they reviewed the incidence of congenital heart disease in 56,109 live births and found 7 cases of vascular rings, which represented 1.5% of all defects.[6]A number of anomalies are associated with vascular rings, mainly the congenital heart disease and is seen in up to 12% of patients.[7]

Aortic development begins in third week of fetal life. When there is normal anatomic development, six pharyngeal arches develop and regress in a cranial to caudal sequence. Any variations in fetal development at different stages can give rise to vascular ring pathologies, either related to aberrant left sided or right sided arch development.[8] A pulmonary artery sling occurs the left artery originates from the right sixth arch instead of the left sixth arch.

The presentation of a child with a vascular ring depends on the degree of compression of the mediastinal structures mainly the trachea and the esophagus. A ring with double aortic arch anatomy has more severe symptoms and thus presents earlier than other vascular rings.[9] The most common gastrointestinal symptom is dysphagia, but this does not occur during early infancy as liquids are tolerated and usually seen when solid foods are introduced in diet.[10] In addition, there can be weight loss and failure to thrive but again, these also present later in childhood.[10]

The diagnostic evaluation of a child with suspected vascular ring should proceed in stepwise fashion. The first investigatios is the chest radiograph which can show the location of the aortic arch. In a child with a normal, left-sided aortic arch, the aortic knob can be identified. In a right aortic arch, the compression on the right side of the trachea can be seen. The diagnostic yield of an esophagram is relatively low, but it can exclude other potential etiologies, like aspiration or tracheoesophageal fistula.[9]Next investigation is Echocardiography during the work-up for vascular rings, which can detect associated congenital heart disease which are seen in nearly 12% of patients with vascular ring .[7] But limitation of echocardiogram is that ring segments without lumens may not be seen.[10]

CT or magnetic resonance imaging(Cross-sectional imaging) is the imaging modality of choice for the diagnostic evaluation and preoperative planning for these patients with a potential vascular ring.[11,12] can produce High-quality images can be produced by three-dimensional reconstruction of the CT which can accurately identify the vascular anatomy and degree of airway compression.[13] Bronchoscopy can be helpful in some patients who first present with respiratory symptoms, as it can provide useful information regarding the location and degree of airway compression and also for evaluation of pulmonary artery sling in order to find out an associated complete tracheal ring.[10,14]Vascular rings are increasingly diagnosed on prenatal imaging, but the clinical significance of this is only beginning to be understood.

MATERIALS AND METHODS:

The study was primarily a retrospective study of patients with vascular ring who underwent surgery at our institution. This study was conducted at ABIVIMS and Dr RML Hospital, Delhi. The study included patients who were operated on at our institution from 2017 to 2023.

The data was collected during follow up visits on opd basis and also telephonically.The data on clinical observations and radiological findings were extracted from medical charts. The type of management and clinical outcome were also documented. The parents were informed that their participation was voluntary and that the research was anonymous and confidential. The questions and clinical signs, such as tachypnea, wheezing, and stridor, were discussed and explained to the parents/caregivers. The data about the outcomes were extracted as a combination of the reported symptoms by the parents and growth charts.

The results of the performed radiological studies such as the chest radiographs, chest computed tomography-angiography (CTA), and barium esophagography were collected.

Statistical analysis was limited by the small sample size, which is anticipated in rare diseases. The results as descriptive statistics, such as frequencies and percentages, and medians.

Clinical Presentation and Diagnosis:

At the time of operation, 81.8% (9 out of 11) of patients were younger than 1 year and 18.2% (2 out of 11) were older than 3 years. Nine patients (81.8%) were boys. DAA was present in 3, RAA with ALSCA and LLA in 5, and LAA with ARSA in 1 and LPA sling in 2 patients.

Associated Anomalies

Associated extracardiac anomalies were trisomy 21 in 9% (1 of 11) of patients, . Concomitant congenital heart defects were found in 4 patients (36.3%), including tetralogy of Fallot in 3 patient , ventricular septal defect in 1, and atrial septal defect in 1.

Table 1:Classification of Vascular Rings

Vascular R type	No.(%)
Double Aortic Arch	3(27)
Right Dominant	1(9)
Left Dominant	2(18)
RAA-ALSCA,LLA	5(45)
LAA-ARSCA	1(9)
LPA Sling	2(18)

ALSCA, aberrant left subclavian artery; ARSCA, aberrant right subclavian artery; LLA, left ligamentum arteriosum; No., number; RAA, right aortic arch.

Table 2 :Patient characteristics .

Demographics	Patients(N=11)
Mean Weight(kg) at Repair	5.8 kg(3-12 kg)
Mean Age at Repair	13.9months(3-39 months)
Male sex	9(81%)
Concomitant Genetic Syndrome	**n(%)**
Trisomy 21	1(9)
Concomitant Cardiac Disease	n(%)
Tetralogy of Fallot	3(27)
Atrial Septal Defect	1(9)
Ventricular Septal Defect	1(9)

Patients at the time of diagnosis may present with nonspecific symptoms or may present as an incidental finding with associated cardiac diseases. They may present with respiratory or gastrointestinal symptoms. The severity of respiratory symptoms at clinical presentation varied depending on the degree of tracheal narrowing. Respiratory symptoms reported by parents at time of clinical evaluation were noisy breathing in 36% (4 of 11) of patients, stridor in 27% (3 of 11), persistent cough in 36% (4 of 11), shortness of breath in 18% (2 of 11), wheezing in 18% (2 of 11), and recurrent upper respiratory tract infections in 18% (2 of 11). Cyanosis was observed in 18% (2 of 11) of patients.

Gastrointestinal symptoms, such as dysphagia, were found in 27% (3 of 11) of patients, and choking from solid food ingestion was reported by parents in 9% (1 of 11) of patients. Table 3 summarizes respiratory and gastrointestinal symptoms.

Table 3: Patient Symptoms at Clinical Presentation

Symptoms	Patients n(%)
Gastrointestinal	
Dysphagia	3(27)
Choking with solid food consumption	1(9)
Emesis	6(54)
Respiratory	
Noisy Breathing	4(36)
Stridor	3(27)
Persistent Cough	4(36)
Shortness of Breath	2(18)
Recurrent Upper Respiratory Tract Infections	2(18)
Wheezing	2(18)
Apnea/Cyanosis	2(18)

Data are presented as n (%).

The Diagnosis was confirmed using various radiological studies such as the chest radiographs, chest computed tomography-angiography (CTA), and barium esophagography Gastrograffin meal follow through (fig 1).

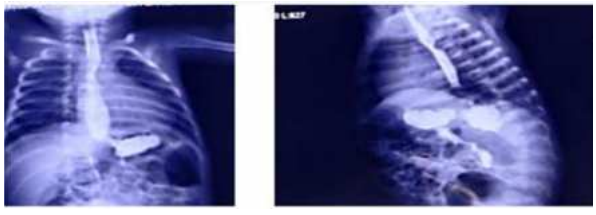

Figure 1: Gastrograffin meal follow through showing indentation on posterior esophagus

Operative Management:

There are four main categories of vascular ring : double aortic arch, right arch with left ligamentum, innominate artery compression, and pulmonary artery sling.

LPA Sling:

First case was a 4 months old male child, weighing 5 kg, was admitted with complaints of frequent chest infections, dyspnoea and stridor . Second case was Echocardiography revealed 5.5mm ostium secundum Atrial septal defect (ASD) and uncertain pulmonary confluence. Cardiac CT was revealed Left Pulmonary Artery (LPA) sling with Left Ligamentum (LL), with tracheal stenosis. Second case was 6 months old male child with LPA sling and mild Tracheal stenosis

Operative Procedure: Operative procedure used was same in both patients. Midline sternotomy was done. LPA was arising from Right Pulmonary Artery (RPA) and traversing behind trachea causing compression. Cardiopulmonary bypass was instituted using aortobicaval cannulation and heart arrested. Direct closure of ASD was done in first case along with ductus arteriosus ligation and division . This was followed by LPA to Main Pulmonary Artery (MPA) translocation. Post-operative recovery was uneventful in both cases and no signs and symptoms of recurrence seen in either patient.

LAA with abberent Rt Subclavian Artery:

6 Months old male child, presented with history of recurrent regurgitation of ingested milk and failure thrive since the age of 1.5 months. Gastrograffin meal follow through was suggestive of posterior indentation of the oesophagus at D4 . CECT thorax and CT Angiography revealed left sided aortic arch with ARSA (distal to origin of left SCA as last branch of arch of aorta) which while crossing the midline is causing posterior compression on oesophagus..

Operative Procedure: Upper partial midline sternotomy done .Innominate vein was identified, looped and retracted inferiorly. Branches of arch of aorta was identified and looped above the innominate vein. Distal right Subclavian artery was identified in the right side in cervical area along which dissection was done till the midline till it was passing behind the esophagus on the right side. The length of dissected ARSA appeared adequate to reanastomose to Right Common carotid artery. So, it was ligated and divided on right side of the oesophagus. The distal transected end of the right subclavian artery was then reimplanted in an end to side fashion to right common carotid

artery using prolene 7-0 as depicted in Figure 2. The pericardium over the arch of aorta was opened between two stay sutures and aortic arch was retracted towards left and anteriorly. Dissection plane was created between left of esophagus and arch and between left of esophagus and descending thoracic aorta (DTA). Proximal ARSA stump was looped and pulled anteriorly left to the oesophagus where it was again ligated at the origin from the DTA.

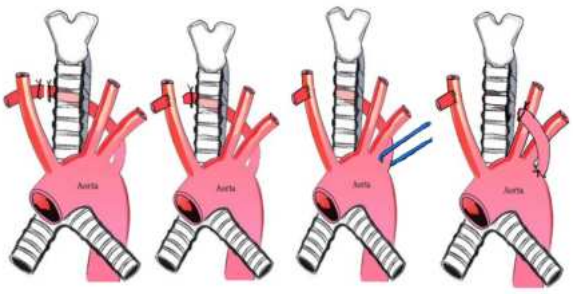

Figure 2: Showing division of ARSCA and its re-implantation on RCCA.

(ARSCA-Abberent Rt Subclavian Artery,RCCA-Rt common Carotid Artery)

Single midline approach for this condition is rarely reported with most cases done by 2 different incisions which increases the post-operative morbidity.Other advantage of this approach is that it gives choices for reimplantation viz ascending aorta, arch of aorta, right common carotid artery.

RAA with Left Ligamentum with ALSCA:

Asymptomatic patient with Vascular ring as an incidental finding:

There were total 5 patients with RAA with Left Ligamentum with ALSCA, out of which 3 were asymptomatic who presented with other cardiac defects like TOF and VSD.In these patients, vascular ring was an incidental finding. Two patients were cases of TOF, who presented with cynosis and no obstructive symptoms of respiratory or gastrointestinal system.One patient was a case of VSD who presented with history of recurrent respiratory tract infections but no obstructive symptoms.

Operative Procedure:

Midline sternotomy was done.Ligamentum arteriosum was dissected,ligated and divided. In all 3 patients,ALSCA was arising distal to origin of RSCA and passing behind esophagus and trachea. In two of three patients ,ALSCA was arising from Kommerell's diverticulum .ALSCA was left in situ in all three patients without any re-implantation as there this was causing no tracheal or esophageal obstruction.Cardiopulmonary bypass was instituted using aortobicaval cannulation and Intracardiac repair was done. Post-operative recovery was uneventful in all cases and there were no signs and symptoms of obstruction on midterm follow up(3-5 yrs) of these patients .

Symptomatic patients with RAA with ALSCA:

1. Lt Thoracotomy Approach:

A 3 months old male child weighing 3 kg who presented with symptoms of cough,dyspnea and strider since 1st week of life, chest x-ray was done which was suggestive of RAA and mild tracheal deviation towards left.CT Cardiac Angiogram revealed RAA with ALSCAarising from Kommerell's diverticulum and Left Ligamentum arising from tip of diverticulum and joining LPA origin thus forming a complete ring causing esophageal and lower tracheal compression.

Operative Procedure: A left thoracotomy approach was used. LL was divided. After dissection of fibrous tissue, it was found that Kommerrel's diverticulum was away from non vascular structures, so instead of the conventional approach of division of diverticulum and re-implantation of LSCA ,diverticulopexy was done to left rib.Post-operative recovery was uneventful and patient is symptom free at 3 years follow-up.

2. Midline Sternotomy Approach:

A 8 Months old male child, presented with history of recurrent regurgitation of ingested milk and failure thrive since the age of 1.5 months. CT cardiac Angiography was done which showed high right sided aortic arch with 4 vessel branching with ALSA with Kommerells diverticulum and atretic ligamentum arteriosus with formation of vascular ring causing compression on oesophagus.

Operative Procedure: Midline sternotomy was done .Branches of arch of aorta and Descending aorta were identified and looped above the innominate vein. 2 vascular clamps applied at the bulbous enlargement of aberrant Left SCA at the origin and divided between the clamps. The distal transected end of the left subclavian artery was then reimplanted in an end to side fashion to left common carotid artery using prolene 6-0. Proximal end of the diverticulum closed with 6-0 prolene continuous suture.Post-operative recovery was uneventful and there are no residual or recurrent symptoms at 1 years follow-up.

This single midline approach for this condition reduces the post-operative morbidity as compared to 2 incision approach which is conventionally used.

Double Aortic Arch:

DAA with Lt Dominant Arch with Lt PDA and Lt DTA :

A 6 months old male child weighing 4kg, presented with dyspnea, dysphagia and repeated hospitalizations since second week of life. Bronchoscopy revealed near total lower tracheal narrowing around 1.5cm above carina. Cardiac CT angiogram confirmed left dominant DAA with left DTA and left patent ductus arteriosus (PDA). Right arch was found looping behind the esophagus with Right Common Carotid Artery (RCCA) and Right Subclavian Artery (RSCA) arising from proximal right arch. Thus leading to tracheo-oesophageal compression.

2nd case with DAA was a 7 months old male child weighing 4.1 kg who presented with severe pneumonia. Cardiac CT angiogram showed left dominant DAA with left DTA , patent ductus arteriosus (PDA) and lower tracheal narrowing.

Operative Procedure: Left postero-lateral thoracotomy was done in both cases. PDA was looped, ligated and divided. Both arches and LSCA were looped . Right arch was doubly clamped proximal to its confluence with left arch and divided, Both its ends were over sewed with double layer prolene sutures in continuous manner. Fibrous tissue around trachea and esophagus was dissected and divided.Both Patients are aymptomatic at 2 yrs follow up.

DAA with Rt Dominant Arch With ALSCA with TOF

A 3 year old female child presented with tachypnea and central cyanosis. Chest X-ray showed tracheal indentations along with features suggestive of Tetralogy of Fallot (TOF). Echocardiography conrmed the diagnoses of TOF with DAA and bilateral superior vena cavae. CT Angiography confirmed Rt dominant DAA with Rt DTA causing Tracheo-esophageal compression.

Operative Procedure: After looping proximal left arch and proximal left subclavian artery, ductus was divided. Cardiopulmonary bypass was instituted using aortobicaval cannulation. The length of left sided distal arch accessible from this approach for division was short, so we performed LSCA blalocking maneuver. In this technique, after doubly snaring the subclavian artery, the arch clamp was shifted proximal to the left subclavian origin. Then the arch distal to LSCA was divided and ends oversewed with prolene double layer running sutures. Fibrous tissue around trachea and oesophagus was dissected and divided. Left superior vena cava was then cannulated and Intra-cardiac repair for TOF was done.

DISCUSSION :

Vascular rings, including double aortic arch and right aortic arch with aberrant left subclavian and left ligamentum, are part of a larger group of vascular-related aerodigestive compression syndromes that also includes aortic arch anomalies,innominate artery compression syndrome, dysphagia lusoria and aneurysms of either the aorta or pulmonary artery. Vascular rings can be isolated or may be in association with other congenital heart disease that is seen in up to 12% of patients.[7]

In this study, we report the the outcome of individualized approach to the diagnosis and management of these syndromes. The presentation of a child with a vascular ring depends on the degree of compression of the mediastinal structures mainly the trachea and the esophagus.

Chest X-rays may be helpful when the right-sided aortic arch indents the trachea. Echocardiography is useful to identify other associated cardiac anomalies but has lower accuracy in patients with double aortic arch and atresia of one of its branches.[15]Contrast swallow studies can also be helpful. The lateral view demonstrates a posterior indentation caused by the encircling posterior arch[16]. Another investigation that can be useful is Flexible bronchoscopy which suggest the diagnosis by finding the typical tracheal compression .[17] The diagnosis is confirmed by CTA, which also defines the anatomy for surgical planning .[16]

This study assessed the outcomes of personalised approach of vascular ring repair in terms of surgical approach either Midline Sternotomy or two incision approach or Thoracotomy, showing comparable results in terms of surgical exposure and better outcomes in terms of decreased postoperative morbidity using single midline sternotomy incision.In this study ,we also assessed the outcomes of different surgical techniques used like complete division of vascular ring or diverticulopexy in symptomatic patients, and only dividing ligamentum arteriosum with repair of other associated cardiac anomaly like TOF or VSD in asymptomatic patients where vascular ring is a incidental finding.On midterm follow-up in these patient ,we found no significant difference in different surgical techniques used in terms of residual or recurrent respiratory or gastrointestinal obstructive symptoms. Freedom from reoperation after vascular ring repair was 100% at 3-5 years . 3 patients who only underwent resection of ligamentum arteriosum and no diverticulum resection or aberrant left subclavian artery reimplantation at the time of surgery ,were also free from reintervention. Thus ,we can use a patient personalised approach for vascular ring repair for better surgical and postoperative outcomes.

Limitations of the study

Our study reported a small sample size. There is a recall bias given the retrospective design of the study.

CONCLUSIONS :

vascular ring include congenital aortic arch anomalies that lead to compression of the trachea, or esophagus, or both. A standardized approach to diagnostic evaluation allows for adequate operative planning. Due to the broad spectrum of disease, the surgeon have a variety of techniques available to address each distinct feature and surgical approach can also be individualised. Single midline Sternotomy incision provide as good exposure as conventional double incision approach and better post operative outcome in terms of decreased morbidity than the double incision approach.

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