Chapter

44 Vascular Ring

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Case Scenario

A 14-month-old boy weighing 8 kg with a history of frequent upper respiratory tract infections and gastroesophageal reflux presented to his pediatrician for a well-child visit. He has not been gaining weight and his reflux seems to be worsening, with frequent postprandial fussiness and increasingly frequent vomiting noted by his parents, especially with solid foods. Although he has always been a noisy breather, his parents have recently noticed increased wheezing with exertion. His pediatrician suspected a vascular ring and ordered a chest radiograph that demonstrated a right-sided aortic arch. The patient was referred to a pediatric cardiologist who is now requesting a computed tomography scan for anatomic delineation and potential surgical planning. Due to his respiratory symptoms, direct laryngoscopy and rigid bronchoscopy are also planned and will be performed in the operating room. These procedures have been scheduled as a single anesthetic, with a transthoracic echocardiogram to be performed in the post-anesthesia care unit while the patient is still sleepy.

Key Objectives

- Understand the anatomic implications of a vascular ring and the most common types.
- Describe anesthetic induction and management strategies for a patient with an unrepaired vascular ring for computed tomography scan and bronchoscopy.
- Understand the surgical approach for repair of vascular rings.

Pathophysiology

What is a vascular ring and what are the anatomic implications?

A vascular ring is a congenital malformation of the aorta and its branch vessels in which the esophagus and/or tracheobronchial tree are either completely ***(complete ring)*** or partially ***(incomplete ring)*** encircled by vascular structures and/or their atretic remnants, resulting in mechanical obstruction of these structures and resultant symptomatology. The term "vascular ring" was first used in 1945 when Robert E. Gross described the first successful surgical repair of a double aortic arch [1]. (See Figure 44.1.)

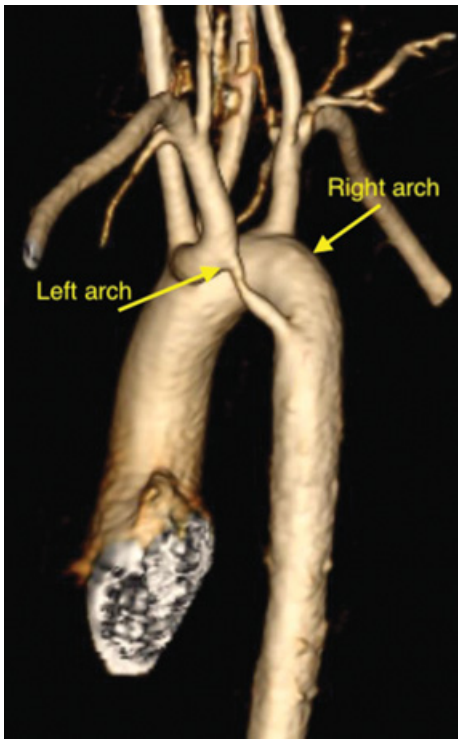

Figure 44.1 Double aortic arch. 3D magnetic resonance imaging of double aortic arch viewed from the left side. There is a dominant right arch and smaller left arch forming a vascular ring. The trachea and esophagus are contained within the ring. Courtesy of Michael Taylor, MD.

How commonly do vascular rings occur?

Vascular rings have a reported incidence of 1/1000 to 1/10,000 births, accounting for roughly 1%–3% of all

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congenital heart disease [2]. Because a large number of vascular rings are asymptomatic, it is likely that the actual prevalence is higher.

What are the most common types of vascular rings?

The most commonly diagnosed types of complete vascular rings are double aortic arch (DAA; 19%–60%) and right aortic arch (RAA) with aberrant left subclavian artery (LSCA)/ligamentum arteriosum (25%–79%), while the most commonly diagnosed types of partial vascular rings are left aortic arch (LAA) with aberrant right subclavian artery (RSCA) (16%–34%), pulmonary artery sling (2%–10%), and anomalous innominate artery (0.54%)[3–6]. The large variance in relative prevalence is reflective of the single-center nature of the published retrospective studies. To improve reporting, the Congenital Heart Surgery Nomenclature and Database Project has subclassified vascular rings as DAA, RAA/left ligamentum, pulmonary artery sling, and innominate compression since the overwhelming majority of vascular rings (>95%) are captured by this nomenclature system [7].

What is the underlying embryopathogenesis of a vascular ring?

The aorta begins to develop during the third week of gestation [7, 8] as blood passes from the endocardial tube through the aortic sac (distal truncus arteriosus) and into the paired dorsal aortae via a series of six paired aortic arches (pharyngeal arch arteries) that sequentially develop and regress in a cranial to caudal fashion [9]. As successive arches regress, portions of the first three are incorporated into the mature vascular structures of the head and neck (first – maxillary artery, second – stapedial and hyoid arteries, third – common and portion of internal carotid), while the fifth either regresses fully or doesn't form at all. The fourth and sixth arches contribute to the definitive aortic arch and the mediastinal segments of the pulmonary arteries as well as the ductus arteriosi, respectively [10]. Vascular rings result from abnormal segmental regression and/or persistence of portions of the embryonic aortic arch complex.

The normal morphology of the definitive aortic arch is left-sided, crossing the left mainstem bronchus at the level of the fifth thoracic vertebra and descending to the left of midline. In DAA, formation of two aortic arches (right and left) occurs due to the persistence of the distal right fourth arch. The rightward more cranially located arch is often dominant and the leftward arch is diminutive or atretic to a varying degree [4]. Each arch gives off their respective common carotid and subclavian arteries. There is usually only one ductus arteriosus, generally located on the left side, and the descending aorta is most commonly located contralateral to the midline from the dominant arch. The right recurrent laryngeal nerve passes around the aorta rather than the RSCA [9].

Clinical Pearl

Vascular rings result from abnormal segmental regression and/or persistence of portions of the embryonic aortic arch complex. In DAA, formation of two aortic arches (right and left) occurs due to the persistence of the distal right fourth arch.

How and when do patients with a vascular ring typically present?

The timing and nature of patient presentation depends on the anatomy of the vascular ring and the degree of the resultant tracheobronchial and/or esophageal compression. Often symptoms are nonspecific, so a high index of suspicion for this diagnosis must be maintained. Most patients will present with respiratory symptoms (88%–95%) with or without esophageal symptoms; isolated digestive symptoms are relatively rare (11%)[11]. (See Figure 44.2.) Infants may present with recurrent upper respiratory infections (URIs) or pneumonia, wheezing, stridor, slow feeding, gastroesophageal reflux disease (GERD), vomiting, and/or failure to thrive. In children and older patients,

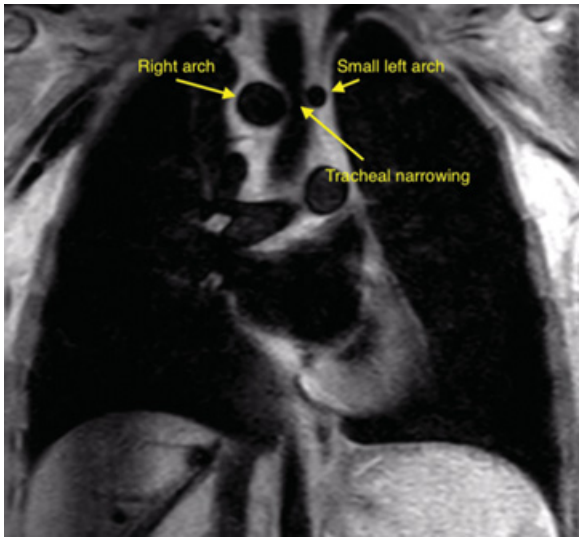

Figure 44.2 Double aortic arch. Coronal black blood magnetic resonance image showing left–right narrowing of the trachea by the double aortic arch. Courtesy of Michael Taylor, MD.

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the symptoms are similar to those seen in infancy with the addition of dysphagia to solids (i.e., dysphagia lusoria secondary to the aberrant RSCA) and dyspnea on exertion.

Clinical Pearl

Tight rings with significant compression, as seen in DAA, may present early in the neonatal period with stridor, wheezing, respiratory distress, and apnea with feeds. Less restrictive rings may present later in life or may remain asymptomatic and be incidentally identified.

Are associated lesions and/or syndromes commonly seen?

While DAA and RAA with aberrant LSCA are isolated anomalies in the majority of cases, the reported incidence of concurrent cardiac lesions for all vascular rings is 12%–32% [3, 5]. The most common associated cardiac defect is tetralogy of Fallot; ventricular septal defect, atrial septal defect, aortic coarctation, transposition of the great arteries and complex single ventricle lesions have also been described [12]. In patients diagnosed with a pulmonary artery sling, coexisting complete tracheal rings can be present in as many as 65% of cases [13].

Aortic arch anomalies, including vascular ring, can be associated with a number of genetic syndromes, most notably 22q11.2 deletions (DiGeorge, CATCH-22, velocardiofacial syndrome, conotruncal anomaly face) and duplications [14]. Trisomy 21, VACTERL (vertebral defects, anal atresia, cardiac defects, tracheoesophageal fistula, renal anomalies, and limb abnormalities) association, CHARGE (coloboma, heart defects, atresia choanae growth retardation, genital abnormalities, and ear abnormalities) syndrome, and PHACE (posterior fossa brain malformations, hemangiomas of the face, arterial anomalies, cardiac anomalies, and eye abnormalities) syndrome have all been reported [14, 15].

How are vascular rings diagnosed?

Vascular rings can be diagnosed via numerous imaging modalities including chest radiography, echocardiography (including fetal), angiography, computed tomography (CT) scanning, magnetic resonance imaging (MRI), barium esophagography, and direct laryngoscopy and bronchoscopy [3, 11, 16]. The choice of imaging strategies is generally based on symptomatology, local expertise, the specialists involved, the availability of advanced imaging technology, and cost. There has been a trend away from barium esophagography and angiography toward CT and MRI, with echocardiography and bronchoscopy also recommended for most patients [10, 11]. In patients diagnosed with a pulmonary artery sling, bronchoscopy should be performed to rule out the presence of complete tracheal rings. Although the optimal diagnostic imaging algorithm for vascular rings is unknown, the goal of imaging should be identification of the relevant vascular and airway anatomy causing the patient's symptoms to minimize unnecessary testing and allow for surgical planning.

How are vascular rings repaired?

Surgical division of a vascular ring is indicated when symptoms of airway and/or esophageal compression are present, and it is generally performed without delay once a diagnosis is made. The approach is dictated by the anatomy of the vascular ring and the presence of any intracardiac and/or tracheal anomalies requiring concurrent repair using cardiopulmonary bypass (CPB). Most commonly, repair is performed via a left posterolateral muscle-sparing thoracotomy (contralateral to the dominant arch) although a right-sided thoracotomy is necessary in the rare case of a left-dominant DAA or right-sided ductus arteriosus. Sternotomy and CPB are utilized for aortic uncrossing procedures for circumflex aorta, for cases when an intracardiac repair is also indicated, and for division/reimplantation of the left pulmonary artery with slide tracheoplasty in the case of complete tracheal rings with pulmonary artery sling. Thoracoscopic and robot-assisted approaches to vascular ring division have been described and are most appropriate in cases where the segment to be divided is not patent [17]. In all cases, the ligamentum arteriosum is divided.

- ***Double aortic arch***: With DAA, the vascular ring is repaired by dividing the most diminutive (or atretic) portion of the nondominant arch. If there is not a clear target for division, it may be necessary for the surgeon to apply test clamps to the potential sites of division to assess for limb pressure gradients (there should be no pressure gradient with occlusion).
- ***Right aortic arch with aberrant LSCA***: Classically, the surgery for RAA with aberrant LSCA is division of the ligamentum. In most patients with this anomaly, the LSCA arises from a remnant of the left fourth dorsal arch called the retroesophageal diverticulum of Kommerell. Increasingly, surgeons are resecting this at the time of vascular ring division and reimplanting the LSCA on the left carotid artery with the goal of preventing residual esophageal compression and the risk of late rupture [18].

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Anesthetic Implications

What are the perioperative considerations for patients with unrepaired vascular rings undergoing diagnostic evaluation?

Patients with a suspected or known (but unrepaired) vascular ring that are undergoing diagnostic evaluation pose a challenge to the anesthesiologist. A detailed history and physical should be conducted with special attention paid to the cardiac, respiratory, and gastrointestinal systems. An overwhelming majority of patients with unrepaired vascular rings will present with some degree of obstructive airway symptoms. The type, severity, and time course of respiratory symptoms (including recurrent infections) should be established, as should the patient's current respiratory status and degree of optimization. Often, the anatomic nature and degree of the underlying airway obstruction are not known during the patient's initial workup. It should be appreciated that signs and symptoms from a concurrent cardiac abnormality may be masked by those of the patient's respiratory pathophysiology (tachypnea, intercostal retractions, nasal flaring, mild intermittent cyanosis), especially when chronic lung disease is present. Diagnostic procedures should be performed in a care setting that will allow for close postoperative monitoring and the ability to expeditiously escalate the level of care in the event that airway manipulation and/or residual anesthetic effects lead to an acute increase in airway obstruction during the postprocedural period.

Clinical Pearl

Patients with underlying genetic syndromes may have additional airway considerations such as subglottic stenosis or cleft lip/palate. Cardiac lesions should also be screened for given the significant incidence of intracardiac lesions in patients with genetic syndromes.

Should any other evaluation or optimization be considered prior to CT and bronchoscopy in this patient? Should echocardiography be performed first?

Even in the absence of concerning symptoms, it would not be unreasonable for a limited transthoracic echocardiogram to be performed without sedation given the relatively high incidence of concurrent congenital heart disease. The patient should also be optimized from a pulmonary standpoint and URIs or pneumonias aggressively treated. To that end, a pulmonology consultation may be helpful. Because the signs and symptoms of 22q11.2 deletions can be subtle, laboratory screening for hypocalcemia and genetic testing should be strongly suggested in patients with vascular ring and conotruncal lesions.

Clinical Pearl

Symptoms of heart disease that cannot be explained by a respiratory etiology (e.g., murmur, pulmonary edema, hepatosplenomegaly, ascites, or four-extremity blood pressure differential) should prompt a preanesthesia echocardiogram and cardiology consult.

Should the case be delayed if the patient presents with a resolving respiratory infection?

The risks and benefits of proceeding with anesthesia in a patient with a vascular ring and a URI must be carefully weighed. Given the patient's respiratory symptomatology it would be best to allow recovery from an active URI prior to administration of an anesthetic if possible. Undue delays in surgery have the potential to hamper normal airway development and/or lead to progressive airway and lung injury [9].

Clinical Pearl

Although an attempt should be made to maximally optimize the patient's respiratory status ahead of a planned anesthetic, it must be appreciated that the definitive treatment strategy for recurrent infections in these patients is surgical repair of the vascular ring.

Should these procedures be bundled and performed at the same time?

The risk-benefit profile of performing multiple diagnostic procedures, potentially under the same anesthetic, should be approached in a patient-specific, team-based manner.

Important considerations include the following:

- Can adequate CT images be obtained without sedation or anesthesia, avoiding anesthesia in the out-of-operating room (OOR) setting and transport entirely?
- If the patient requires intubation, would it be safer to perform the bronchoscopy and intubation in the operating room (OR) prior to CT, allowing for careful endotracheal tube (ETT) placement proximal to the area of the obstruction, with return to the OR for extubation?

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- If the CT protocol calls for a free-breathing patient, can the patient be safely induced in the OOR setting and then transferred sedated with an unsecured airway to the OR?
- If the patient is not intubated for the CT, can/should the bronchoscopy be delayed and instead performed intraoperatively during the vascular ring repair to avoid transport entirely?

Adequate airway support and vital signs monitoring during procedures in OOR settings and the transport to and from these locations is critical. According to the American Society of Anesthesiologists (ASA) Closed Claim Analysis Project, respiratory events (inadequate oxygenation and ventilation) are the most common type of adverse events in OOR locations and tend to be more severe in nature as compared to in-OR procedures [19]. A preponderance of respiratory events was also described by the Pediatric Sedation Research Consortium's multiple analyses of the adverse events encountered during sedation of pediatric patients in OOR settings [20, 21].

What are the major anesthetic considerations for this patient presenting for CT scan and bronchoscopy?

The overwhelming majority of patients with a vascular ring will present with some degree of respiratory symptoms and airway compromise due to an unknown combination of external obstruction, tracheobronchomalacia, and/or tracheal stenosis. Unfortunately, anesthetic goals for managing these various airway pathologies can be somewhat conflicting.

Specifics of the anesthetic plan will depend on

- The ***degree of contribution of direct airway compression*** and/or secondary airway disease (tracheobronchomalacia, interstitial lung disease) to the patient's symptoms
- The ***imaging protocol*** utilized (the need for breath-holding versus free-breathing during image acquisition)
- The ***logistics of intubating the patient*** in the OR versus an OOR setting

The choice of intravenous (IV) versus inhalation induction should be made after consideration of the severity of the patient's airway and/or gastrointestinal symptomatology; an inhalation induction is generally well tolerated. If the patient requires intubation for either respiratory or logistical reasons, the patient should be induced in a careful manner and the ability to ventilate with positive pressure ventilation should be confirmed prior to the administration of neuromuscular blockade. If necessary, intubation and extubation should be performed in a controlled, familiar setting with adequate technical and material support including additional anesthesia personal, readily available airway equipment (e.g., smaller endotracheal tubes and appropriately sized suction catheters, bronchoscopy for endotracheal tube troubleshooting, noninvasive positive pressure ventilation devices), and rescue medications (e.g., dexamethasone, nebulized racemic epinephrine, albuterol, succinylcholine). If monitored anesthesia care is chosen, the provider should closely monitor oxygenation and ventilation and be prepared to rapidly treat episodes of desaturation, apnea, laryngospasm, and/or excessive airway or pooled oropharyngeal/esophageal secretions. Choices for titrated medications for monitored anesthesia care include midazolam, dexmedetomidine, and/or ketamine.

What anesthetic induction, monitoring, and airway management considerations exist for vascular ring repair?

The preoperative imaging and operative plan (sidedness, temporary or permanent vascular occlusion) should be carefully reviewed. A gentle mask induction with sevoflurane is generally well tolerated if gastrointestinal symptomatology does not preclude this and allows for the performance of bronchoscopy in a spontaneously breathing patient if further airway assessment is required. In addition to recommended standard ASA monitors, the placement of an arterial line is generally indicated, especially in cases when significant aortic manipulation is anticipated.

Clinical Pearl

The location of the noninvasive blood pressure cuff(s) and arterial line should be determined by the patient's anatomy and the type of repair to be undertaken:

- ***DAA***: Bilateral upper extremity and unilateral lower extremity
- ***RAA with aberrant LSCA***: Right radial arterial line if the LSCA is to be translocated or sacrificed
- ***Vascular ring repair with concurrent anterior–posterior tracheopexy***: Upper and lower extremity arterial lines

At a minimum, there should be a way to reliably measure pressures in both an upper and lower extremity. The application of cerebral near-infrared spectroscopy monitors should be considered if vascular clamping is anticipated. A central venous line may be indicated for some cases to allow for the infusion of vasoactive medications and to provide additional access. Although blood loss is generally

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minimal, there is the potential for massive hemorrhage, so vascular access and blood product availability should be planned with this in mind. Transesophageal echocardiography is usually reserved for cases requiring CPB.

What is an appropriate emergence and extubation plan after vascular ring repair? What modalities can be considered for postoperative analgesia?

Most patients undergoing vascular ring division are candidates for early extubation in the absence of significant pulmonary disease, the performance of a complex intracardiac or tracheal repair, and/or significant residual lesions. Enhanced analgesia with neuraxial (thoracic epidural) or peripheral nerve blockade (paravertebral nerve block or erector spinae plane block +/− catheter placement) can aid in facilitating early extubation, as can the use of multimodal analgesia and intravenous acetaminophen.

What are potential immediate postoperative concerns?

Vascular ring division is associated with a very low perioperative mortality rate (0–5%) and a relatively low postoperative complication rate (2%–8%)[5, 6, 11]. Although repair results in the immediate relief of tracheal and esophageal compression in the overwhelming majority of patients, persistent tracheal obstruction due to residual lesions and/or tracheomalacia has been reported in >10% of patients [15]. Recurrent laryngeal nerve paresis/paralysis is seen in up to 8% in some series [11]. Other immediate postoperative concerns include bleeding, chylothorax, and pneumonia.

What is the expected long-term outcome for these patients?

Long-term mortality, freedom from reoperation, and complication rates are excellent with the majority of patients reporting either partial or complete relief of symptoms. In a certain percentage of patients, tracheomalacia does resolve at longer follow-up intervals [6]. Postoperative pulmonary function tests, however, have been shown to continue to demonstrate significant airway obstructive patterns even in asymptomatic patients. As the resection of retroesophageal diverticulum of Kommerell and aggressive surgical management of tracheomalacia become more commonplace at the time of vascular ring repair, the rates of residual and recurrent symptoms, long-term complications, and the need for reoperation may decrease further [22].

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