Bryson A, Carlisle M, Langenfeld TL, Wilhelm CM, Pasternack DM. Vascular Rings in Review: Historical Shifts, Current Trends, and Future Potential. J Pediatrics & Pediatr Med. 2026; 6(4): 1-6

Journal of Pediatrics and Pediatric Medicine

Journal of Pediatrics and Pediatric Medicine

Short Review Article Open Access

Vascular Rings in Review: Historical Shifts, Current Trends, and Future Potential

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Article Info

Article Notes
Received: May 22, 2026
Accepted: June 30, 2026

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Keywords:
Congenital Heart Disease
Vascular Ring
Double Aortic Arch
Computed Tomography
Magnetic Resonance Imaging
Fetal Diagnosis

Abstract

Vascular rings are congenital anomalies of the aortic arch with varying degrees of respiratory and digestive symptoms due to anatomic compression. The most common types are double aortic arch (DAA) and right aortic arch with an aberrant left subclavian artery (RAA-ALSA). Fetal imaging has greatly advanced in the last three decades, leading to increasing prenatal diagnosis of vascular rings with growing operator comfortability and standardization of dedicated cardiac views. Improved early detection has led to earlier surgical intervention, even prior to symptom onset. Cardiac computed tomography (CT) has become the preferred modality for postnatal diagnosis as it provides clear, high-resolution imaging to allow for better surgical planning. The field continues to grow as new technologies and imaging modalities, such as 3D-printing, virtual reality, and four-dimensional fetal ultrasound, are being explored to further improve early and accurate detection of vascular rings.

Introduction

A vascular ring is a congenital anomaly of the aortic arch that causes varying respiratory and digestive symptoms due to anatomic compression. First described in 1737 and first repaired in 1945, the approach to diagnosis and treatment of vascular rings has changed dramatically in recent times[1,2]. In this article, we review the basic principles of vascular rings while providing historical background, insights on current diagnostic trends, and future possibilities.

Clinical Presentation

As discussed in Table 1, the most common forms of vascular ring include double aortic arch (DAA) and right aortic arch with an aberrant left subclavian artery and left-sided ductus arteriosus (RAA-ALSA)[3-7]. A DAA is formed by persistence of the embryonic right and left fourth branchial arches. The ascending aorta bifurcates into two arches that encircle the esophagus and trachea before rejoining as the descending aorta. The right arch commonly gives rise to the right common carotid and right subclavian arteries, while the left arch gives rise to the left common carotid and left subclavian arteries. RAA-ALSA is considered a "looser" ring, with the trachea and esophagus compressed rightward by the right-sided aortic arch and posteriorly by the diverticulum of Kommerell (Figure 1). This is a bulbous dilation at the origin of the aberrant artery extending from the aorta to the insertion of a ductus arteriosus or ligamentum arteriosum. This left-sided ductus or ligamentum completes the ring around the mediastinal contents.

These 3-dimensional reconstructions demonstrate the anatomy of a vascular ring formed by a right aortic arch with aberrant left

Table 1. A description of the most common vascular rings

Characteristic	Double aortic arch	Right aortic arch with aberrant left subclavian artery
Ring anatomy	Left & right arch encircle the mediastinum; 1 arch usually larger (dominant)	Aortic arch on one side of the mediastinum with a contralateral ligamentum arteriosum, completed posteriorly by the aberrant subclavian
Tightness	Tighter	Looser
Surgical treatment	Division of non-dominant aortic arch	Division of ligamentum +/- reimplantation of subclavian

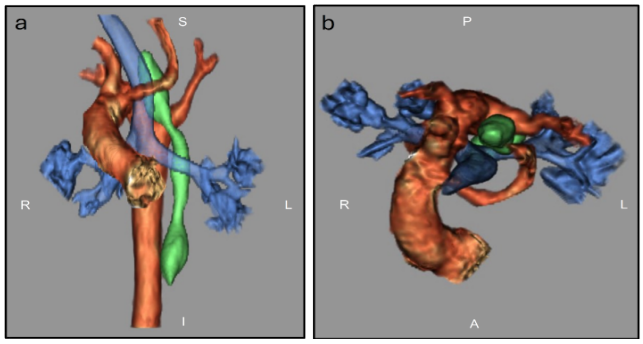

Figure 1. 3-dimensional reconstructions of a right aortic arch with aberrant left subclavian artery
R: right, L: left, S: superior, I: inferior, A: anterior, P: posterior

subclavian artery. (a) the reconstruction from a coronal view. (b) the reconstruction from an axial view.

Multiple other, less common vascular ring configurations have been described, often forming incomplete rings that vary in severity of symptoms. The individual prevalence of each type is not frequently commented on, and given their asymptomatic potential, these numbers are thought to be underestimated. The overall prevalence of vascular rings is 1 in 10,000 live births. The incidence is estimated to be three percent of congenital heart defects and up to two times higher in males[3-5,8,9]. A greater incidence of vascular ring has been described in genetic conditions, classically 22q11 microdeletion syndrome, but also trisomy 21 and CHARGE syndrome.

Symptomatic presentation varies, with the location and severity of compression directly correlating to symptom severity, earlier presentation, and higher likelihood of surgical intervention. The average age at presentation of rings requiring surgical intervention is around one year[3-5,7,10]. Patients with less severe compression may develop symptoms later in life or never at all. Respiratory symptoms included stridor, recurrent infections, respiratory distress, cough, and wheezing. Esophageal symptoms included dysphagia, feeding difficulties, vomiting, poor weight gain, and preference for liquids[4,10-12]. Dysphagia is more common in older children and adolescent presentations[7,12]. Around 80-90% of patients present with respiratory symptoms, while around 40-50% present with gastric symptoms; nearly 30% present with both[4,11]. Isolated vascular rings do not typically present with cardiac exam findings or symptoms. Given their asymptomatic nature, it is often difficult to determine their true prevalence.

Fetal Diagnosis

Rates of prenatally diagnosed vascular rings are steadily increasing as routine anatomy scans with dedicated cardiac views at 18-22 weeks gestation have become standard of care[13-17]. While the introduction of the 4-chamber and outflow tract views led to detection of upwards of 80% of major structural defects[18], it was not until the three-vessel view became more widespread in the early 2000s that detection of arch abnormalities began to increase[19].

Several studies examining trends in rates of prenatal diagnosis of vascular rings have shown a steadily increasing rate of prenatal detection in the last two decades, likely owing to widespread adoption of the three vessel view as standard of care, with current detection rates ranging from 65-78%[13-17]. As prenatal diagnosis has increased, so has the prevalence of RAA-ALSA[13,14,17]. The literature regarding accuracy of anatomy scan and fetal echocardiography in diagnosing vascular rings is limited, however one study demonstrated a positive predictive value of 81% in 68 suspected vascular rings while another found no missed diagnoses in a cohort of 36,679 fetuses who underwent routine prenatal ultrasound[20,21].

Existing literature suggests that prenatal diagnosis may lead to improved postnatal outcomes. A cohort of 190 patients with vascular rings who underwent primary repair, of which 29 were diagnosed prenatally, was examined and prenatally diagnosed patients were found to have undergone surgical intervention at a significantly younger age (13.1 months vs 24 months) with no difference in postoperative complications or length of stay when compared to postnatally diagnosed patients[16]. Another study analyzed similar outcomes in their cohort of 177 patients where 80 were diagnosed prenatally and also found that prenatally diagnosed patients underwent surgical repair at a younger age (7.5 months vs 16 months)[15]. They also found that postnatally diagnosed patients were more likely to have preoperative respiratory symptoms, been prescribed respiratory medications, and been admitted for asthma or reactive airway disease.[15] Further studies similarly showed evidence that patients with prenatal diagnosis of vascular ring are less likely to present with symptoms, require fewer preoperative medications, and have fewer residual symptoms following repair[13,15,16,20,22]. However, existing studies are observational in nature, limiting the strength of the conclusions we can draw from them.

Table 2. A comparison between magnetic resonance imaging (MRI) and computer tomography (CT) scans in the evaluation of patients with a vascular ring

Characteristic	MRI	CT
Radiation exposure	None	Required
Length of test	Long	Short
Anesthesia requirement	Common for infants & young children	Rare
Resolution	Sufficient for structures of interest	High
Airway evaluation	Limited	Preferred modality
Use pattern	Mostly historical, preferred for serial assessment	Generally preferred today

While routine anatomy scans and standard fetal echocardiography have demonstrated good sensitivity and accuracy in detecting vascular rings, there are limitations such as suboptimal fetal positioning and operator dependence. Therefore, new technologies and imaging modalities are being explored to enhance prenatal diagnostic accuracy and planning of postnatal care. Three- and four-dimensional ultrasound with spatio-temporal image correlation, cardiac MRI, 3D-printing, and artificial intelligence have all been used with the goal of producing better images, enhancing prenatal planning, and lessening operator variability[23-27]. While most studies evaluating these modalities examined their utility for congenital heart disease in general, one study reviewed echocardiographic imaging of 30 patients with fetally diagnosed arch anomalies and found 3D spatio-temporal image correlation was more sensitive and accurate than 2D imaging for detecting arch anomalies[26]. These technologies remain promising, though based on limited observational data. Additional studies will be needed to demonstrate their improvement in clinical outcomes.

Although our ability to detect vascular rings by prenatal imaging is improving, the field remains uncertain on how to approach the asymptomatic patient[28]. In fact, many patients with vascular rings remain asymptomatic throughout life without a requirement for intervention, although it remains unclear how to best predict which patients will be symptomatic or not[9,16,20]. In general, a conservative approach to these asymptomatic patients is preferred, although the exact criteria for obtaining additional testing and even referring for surgery remains a point of discussion[9,20,28]. Care should be taken to avoid unnecessary healthcare costs, radiation or anesthesia exposure, or surgical intervention. Future research with prospective, interventional study designs may help provide additional insight regarding which patients benefit from early repair versus those in whom expectant management is the superior approach.

Diagnostic and Cross-Sectional Imaging

Prior to the adoption of cross sectional imaging, vascular rings were diagnosed through a combination of barium esophagography, angiography, and chest radiography with exact anatomy being discovered intraoperatively through dissection and inspection[29]. The advancement of magnetic resonance imaging (MRI) and computer tomography (CT) brought a shift in strategy with anatomical information about exact arch configuration, tracheobronchial compression, and brachiocephalic vessel branching visualized in ways previously not possible through conventional diagnostic methods[30]. Surgical planning was often reactive to intraoperative anatomical discoveries, but with improved preoperative anatomical understanding via cross sectional imaging, surgeons are able to adjust the approach to each patient's unique anatomy[30,31].

The choice between MRI and CT for evaluation of vascular rings remains controversial, with both able to obtain diagnostic-quality images despite their respective pros and cons (Table 2)[32,33]. Cardiac MRI can provide diagnostic-quality imaging of the intrathoracic vasculature without need for radiation, although is a prolonged study requiring breath holds, often resulting in a need for infants and young children to receive anesthesia to yield diagnostic results [6,33,34]. While CT relies on radiation exposure, it is a quicker test, can evaluate the tracheobronchial tree, and can provide higher-resolution images that are easier to process for 3-dimensional or virtual reality modeling[33-35]. Studies comparing CT and MRI found near equivalence in the visualization of vascular ring anatomy[32, 36].

Historically, MRI was the predominant imaging modality for much of the 1990s and 2000s with CT overtaking MRI around the early-to-mid 2010s[37,38]. CT is currently the preferred imaging modality for evaluation of vascular rings, with some centers making explicit statements that they do not use MRI for routine evaluation of vascular rings[28,37-39]. The shift appears to have begun following the adoption of multi detector CT in the 2000s, further advancements in speed of image acquisition, image quality, and decreased dose of radiation[36]. Determining the appropriate imaging modality has thus become patient-specific with factors such as need for sedation, exposure to radiation, the importance of tracheal imaging, and institutional capability[33].

Similar to the shift in prevalence of imaging modalities, society recommendations have evolved over the years as well; In 2010 the American College of Cardiology Foundation Task Force recommended MRI over CT due to absence of radiation exposure[41]. However, by 2015 the Society of

Cardiovascular Computed Tomography describes CT as the optimal diagnostic modality for evaluation of vascular rings including the assessment of vascular anatomy and associated tracheobronchial narrowing[42]. In 2022 the Society for Cardiovascular Magnetic Resonance Imaging recommended MRI over CT depending on institutional expertise[43].

As imaging continues to evolve, it prompts the question of which emerging modalities may influence the approach to vascular rings. A 2024 scientific statement in the Journal of the American College of Cardiology discussed the emergence and growing application of advanced digital technologies including extended reality, digital twins, and artificial intelligence, concepts that can readily be applied to the evaluation of vascular rings[44]. Newer technologies such as 3D printing and modeling are already impacting the approach towards vascular rings in their ability to reveal findings not detected on conventional image reconstruction[23]. Other modalities such as extended and virtual reality have shown benefit in surgical planning and anatomy education respectively[45,46]. While promising, these emerging modalities and their evolving applications are still investigational, and remain a future opportunity to explore.

Treatment

As a mechanical problem, the treatment of vascular rings is invariably surgical. There is disagreement on the indications for this surgery, although most cardiologists and surgeons reserve surgery for symptomatic patients[9,33].

The detailed approach to surgical correction of vascular rings is outside the scope of this review and varies based on the patient's anatomy. In general, the goal is to achieve a "release" of the ring, usually through ligation and division of a component structure, often the ligamentum arteriosum or an atretic component of a double aortic arch[1]. Some surgeons also perform additional interventions such as aortopexy or reimplantation of involved structures to further reduce the likelihood of tracheoesophageal obstruction[34.] Rarely tracheal intervention may be required[1].

Patients are expected to tolerate surgical release of vascular rings well without many long-term complications, although care must be taken by the surgeon to avoid injury to surrounding structures[34]. While symptoms usually improve post-operatively, persistence of symptoms may occur, especially with "tighter" rings and older age at surgery[34].

Conclusion

Over the years, technological advancements have improved our approach to vascular rings. Using fetal imaging, patients are now diagnosed before they are symptomatic, possibly even preventing symptoms. CT has replaced former imaging modalities to provide a clearer image of the ring while also assessing the airway. The future remains bright, as the field continues to incorporate new and emerging technologies to help provide earlier and more accurate diagnosis.

Acknowledgement

No external financial support, grants, or funding were used for the creation of this manuscript.

Conflict of Interest

No authors have a conflict of interest that would bias findings or conclusions in this manuscript.

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