Vascular Rings and Slings

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Abstract

Since the mid-1940s, the phrase "vascular ring" has referred to congenital vascular anomalies of the aortic arch system that encircle and compress the esophagus and trachea causing various symptoms. The concept of vascular sling was described five decades later. The diagnosis of vascular ring or pulmonary artery sling should be suspected in any infant or child presenting with symptoms of respiratory distress, noisy breathing, or dysphagia. Suspicion of the diagnosis is often generated by the plain chest x-ray. However, the diagnosis is best established by CT imaging which accurately delineates the anatomy of the vascular ring and the associated tracheal pathology. All patients diagnosed with a vascular ring should have an echocardiogram because of the incidence of associated congenital heart disease.

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Bronchoscopy should be performed in all cases to assess for additional tracheal pathology and to provide a clinical assessment of the degree of tracheomalacia and bronchomalacia. This chapter aims to overview the embryology, the anatomy and pathophysiology, the clinical presentation and diagnosis, and the management of the entities included in the spectrum of vascular rings and slings.

Keywords

Aberrant right subclavian artery • Anomalies • Aortic arch syndrome • Complete tracheal rings • Congenital • Double aortic arch • Esophageal stenosis • Innominate artery compression syndrome • Pulmonary artery • Pulmonary artery sling • Right aortic arch • Tracheal stenosis • Vascular malformations

Introduction

The phrase "vascular ring" was coined by Robert Gross in his classic paper in the *New England Journal of Medicine* from 1945 [1]. At that time, Gross described the first successful division of a double aortic arch. Within that article, Gross stated the following:

> A ring of blood vessels was found encircling the intrathoracic portion of the esophagus and trachea... The pathologic findings at once suggested that a division of some part of the *vascular ring* during life would have relieved the pressure of the constricted trachea and esophagus.

Since that time, the phrase "vascular ring" has referred to congenital vascular anomalies of the aortic arch system that encircle and compress the esophagus and trachea causing various symptoms. Figure 123.1 is an autopsy photograph of a child who died with a double aortic arch. The compression of the trachea and esophagus is quite apparent. In the current era, a child such as this would be diagnosed with a computed tomographic angiogram (CTA) as shown in Fig. 123.2. The ability in the current era to define the anatomy so precisely is a great aid to the surgeon in managing these patients. Gross was also the first to describe the other classic vascular ring, that is, the right aortic arch with retroesophageal left subclavian artery and left ligamentum arteriosum [1].

The surgical experience at Ann & Robert H. Lurie Children's Hospital of Chicago began with a case report by Willis J. Potts published in the *Archives of Surgery* in 1948 [2]. Potts reported two patients with double aortic arch. Potts also was the first to report successful repair of pulmonary artery sling [3]. This is a rare vascular anomaly where the left pulmonary artery originates from the right pulmonary artery and encircles the distal trachea en route to the left lung. In 1954, Potts reported intraoperative diagnosis and repair of a pulmonary artery sling in a 5-month-old infant with intermittent attacks of dyspnea and cyanosis. The repair was done through a right thoracotomy utilizing the Potts ductus clamps. Beginning with those two case reports, the series of children undergoing an operation for a vascular ring or pulmonary artery sling at Lurie Children's has grown dramatically as illustrated in Fig. 123.3. The total experience with double aortic arch, right aortic arch, and pulmonary artery sling is shown in Table 123.1. This experience forms the basis for this chapter. The classification of vascular rings used at Lurie Children's is based on anatomic and clinical features of the patients, in particular the location of the aortic arch(es). This classification scheme has been endorsed by the International Congenital Heart Surgery Nomenclature and Database Project for the Society of Thoracic Surgeons [4].

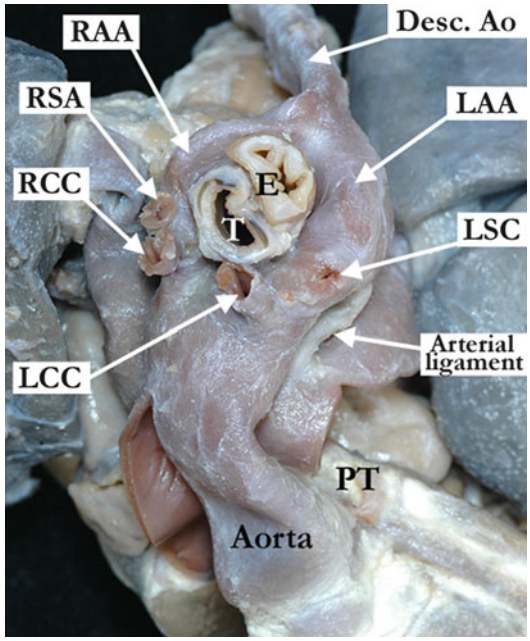

Fig. 123.1 Autopsy photograph of a child who died from airway obstruction who had a double aortic arch. *Desc Ao* descending aorta, *E* esophagus, *LAA* left aortic arch, *LCC* left common carotid, *LSC* left subclavian, *PT* pulmonary trunk, *RAA* right aortic arch, *RSA* right subclavian artery, *RCC* right common carotid, *T* trachea

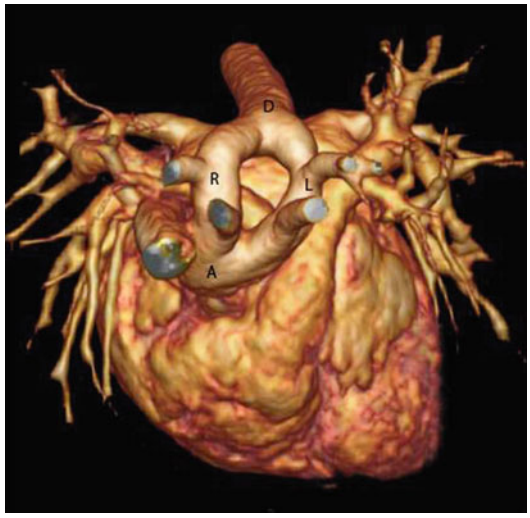

Fig. 123.2 Computed tomogram (3D reconstruction) of a child with a double aortic arch. Same view as in Fig. 123.1. This child has balanced right and left arches. *A* ascending aorta, *D* descending aorta, *L* left arch, *R* right arch

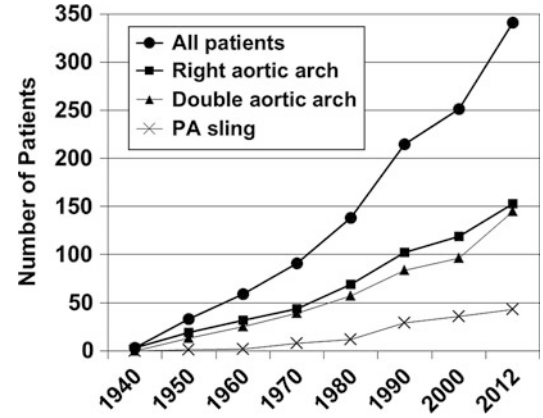

Fig. 123.3 Vascular ring and pulmonary artery sling patients undergoing repair at Ann & Robert H. Lurie Children's Hospital of Chicago

Table 123.1 Lurie Children's experience (1947–2012)

Vascular ring	# of patients
Double aortic arch	145
Right aortic arch/left ligamentum	153
Pulmonary artery sling	43
Total	341

Clinical Presentation and Diagnosis

The classic presentation of a child with a symptomatic vascular ring is noisy breathing and a "seal bark" cough. Other frequent symptoms are wheezing, recurrent upper respiratory tract infections, dyspnea on exertion, and dysphagia. Apnea and apparent life-threatening events can also occur. Some children may have severe respiratory distress requiring intubation and ventilation. Dysphagia usually occurs in older children and is mostly a problem when taking solid foods. A classic symptom is that the child (because they have learned to chew their food very carefully) is the last to leave the table at dinner time. A table of symptoms leading to clinical presentation is shown in Table 123.2.

The diagnostic evaluation of a patient with a vascular ring should proceed in a stepwise fashion until adequate information has been obtained

Table 123.2 Symptoms leading to clinical presentation in patients with vascular rings[a]

	Double aortic arch (n = 80)[b]	Right aortic arch (n = 78)[b]
Stridor	46 (57%)	18 (23%)
Recurrent upper respiratory tract infections	22 (27%)	18 (23%)
Cough	17 (21%)	8 (10%)
Dysphagia	12 (15%)	12 (15%)
Respiratory distress	8 (10%)	13 (17%)
Ventilator preoperatively	7 (9%)	3 (4%)

From Backer et al. [5]
[a]More than one symptom occurred in many patients
[b]Our records did not provide symptoms for the earlier patients in the series

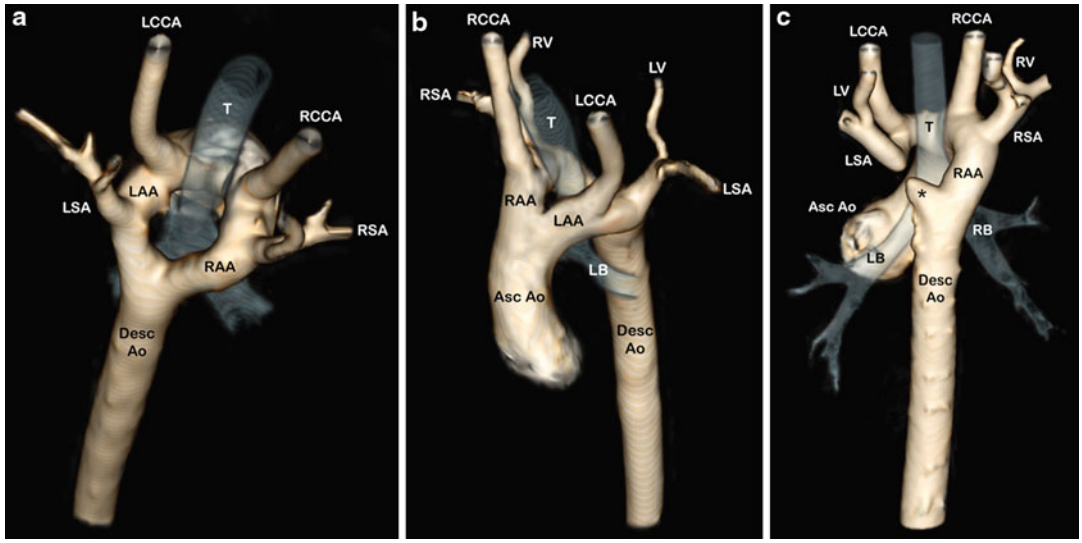

Fig. 123.4 (**a–c**) Contrast-enhanced CT angiograms with 3D multiplanar reformat of children with a double aortic arch. (**a**) Posterior view of balanced arches, (**b**) sagittal oblique view (same patient), and (**c**) posterior view of dominant right aortic arch with left arch atretic between the left subclavian artery takeoff and a small diverticulum (*asterisk) that extends off of the descending aorta. *Asc Ao* ascending aorta, *Desc Ao* descending aorta, *LAA* left aortic arch, *LB* left bronchus, *LCCA* left common carotid artery, *LSA* left subclavian artery, *LV* left vertebral, *RAA* right aortic arch, *RB* right bronchus, *RCCA* right common carotid artery, *RSA* right subclavian artery, *RV* right vertebral, *T* trachea

for the surgeon to intervene. These authors' diagnostic strategies have changed significantly over the past decade. The primary means of diagnosis in the current era is computed tomography (CT) [6]. With the newest generation of dual source CTs the evaluation can be completed in less than 1 s without the need for intubation and with a considerable reduction in radiation dose. The information obtained from a CT allows for precise planning of the surgical strategy. Examples of CT are shown in Figs. 123.4a–c (double aortic arch) and 123.5a, b (right arch).

The use of computed tomography has replaced barium swallow (Fig. 123.6) as the primary means of diagnosis. Some centers prefer magnetic resonance imaging (MRI), but this requires a longer time period to obtain and often needs sedation and intubation. MRI also does not give as clear a picture of the tracheal lumen. Other examinations that can lead to a strong suspicion

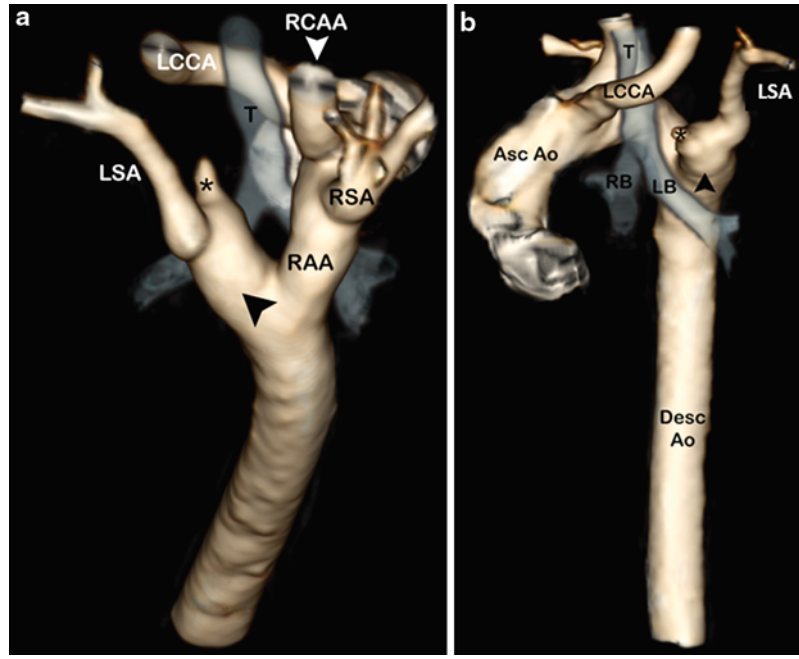

Fig. 123.5 (**a**, **b**) CTA with 3-dimensional multiplanar reformat (MPR) views: (a) posterior view of right aortic arch with aberrant left subclavian artery arising from a diverticulum of Kommerell (*arrowhead*) and ligamentum arteriosum (*asterisk*) and (**b**) MPR view of same patient shown in Fig. 123.5 (**a**). *Asc Ao* ascending aorta, *Desc Ao* descending aorta, *LB* left bronchus, *LCCA* left common carotid artery, *LSA* left subclavian artery, *RAA* right aortic arch, RB right bronchus, *RCCA* right common carotid artery, *RSA* right subclavian artery, *T* trachea

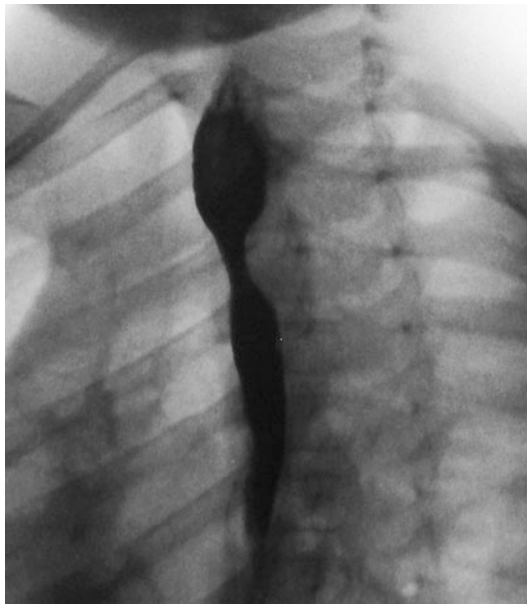

Fig. 123.6 Sagittal oblique view esophagram of a 2-year-old child with circumflex aorta. Note the indentations on the anterior and posterior aspects of the esophagus from the right aortic arch crossing posteriorly to the left

of a vascular ring include chest radiograph, bronchoscopy, and echocardiogram.

The chest radiograph is the most common place to start the evaluation. The chest radiograph will establish the location of the aortic arch whether it is a normal left aortic arch, a right aortic arch, or an indeterminate aortic arch (likely a double aortic arch). Some children who present primarily with noisy breathing or chronic cough will undergo bronchoscopy as their first examination. Bronchoscopic examination will show an extrinsic, often teardrop-shaped (often pulsatile) compression of the trachea. This leads to the suspicion of a vascular ring which can be confirmed by computed tomography. Initially, echocardiography was believed to be an excellent way to diagnose vascular rings. However, because structures that do not have blood passing through them are not visible with echocardiography, this has not become as important a diagnostic tool as was originally thought. However, an echocardiogram is highly recommended in all patients diagnosed

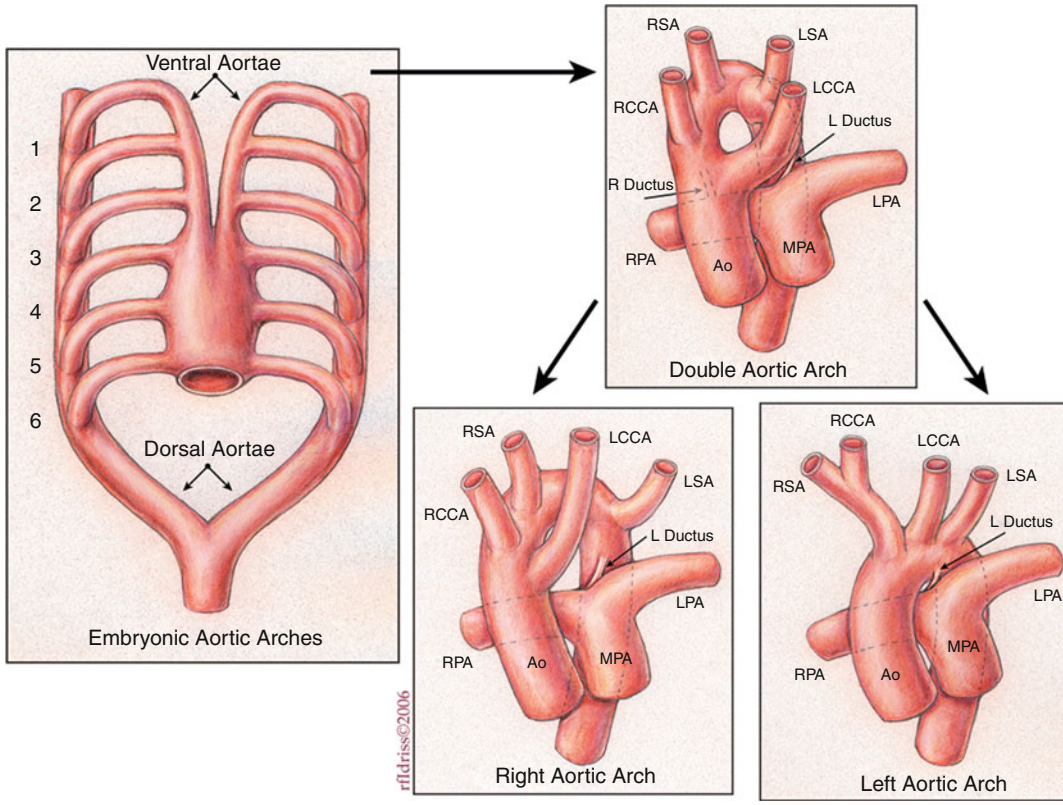

Fig. 123.7 Embryonic aortic arch development. Six pairs of aortic arches originally develop between the dorsal and ventral aorta. The first, second, and fifth arches regress. Preservation or deletion of different segments of the remaining arches results in either a double aortic arch, a right aortic arch, or the "normal" left aortic arch. *Ao* aorta, *CCA* common carotid artery, *L* left, *PA* pulmonary artery, *R* right, *SA* subclavian artery (Reprinted with permission from Backer CL, Mavroudis C, Stewart RD, Holinger LD. Congenital anomalies: vascular rings. In: Patterson GA, Cooper JD, Deslauriers J, Lerut AE, Luketich JD, Rice TW (eds): Pearson's Thoracic and Esophageal Surgery, Philadelphia, Church Livingstone Elsevier, 2008, 242–255)

with a vascular ring because of the 12 % incidence of associated cardiac pathology [5].

Embryology

The embryologic origin of vascular rings depends on varying involutions, regressions, and persistence of components of the embryonic arch system. The embryonic aortic arch system consists of a ventral and dorsal aorta connected by six primitive aortic arches (Fig. 123.7). In the development of the aortic arch, the first, second, and fifth aortic arches initially involute to form Edwards' classic aortic arch [7]. If the arch development arrests here, the patient will have a double aortic arch, a type of vascular ring. If the right fourth arch involutes, a normal left aortic arch is formed and the patient will not have a vascular ring. If the left fourth arch involutes and the right aortic arch remains patent, the patient will have a vascular ring malformation now referred to as a right aortic arch, retroesophageal left subclavian artery, and left ligamentum. There can be a great deal of variation within these major groupings depending on the positioning of the right and left carotid arteries, right and left subclavian arteries, and the location of the patent ductus arteriosus which later becomes the ligamentum arteriosum. As mentioned in the "Clinical Presentation and Diagnosis" section of this chapter, a number of these patients will have associated anomalies. Of particular importance is the 12 % incidence of

associated cardiac pathology. However, the most important association is that between pulmonary artery sling and tracheal stenosis secondary to complete cartilaginous tracheal rings. This is called the ring-sling complex [8]. In patients diagnosed with a pulmonary artery sling, two-thirds will have a tracheal stenosis caused by congenital cartilage tracheal rings. If starting with a diagnosis of tracheal stenosis, fully one-third of those patients will be found later to have a pulmonary artery sling.

Indications and Surgical Technique

The great majority of patients with a vascular ring will have clinical symptoms. Patients with clinical symptoms and a vascular ring or pulmonary artery sling require surgical intervention. The repair should be performed at the time of diagnosis to help avoid serious complications that occur from apnea, hypoxic episodes, or severe respiratory upper tract infections requiring intensive care unit admission. Other reported complications from unrepaired vascular rings include aortic dissection, aortic aneurysm, and catastrophic bleeding that can occur when patients have indwelling nasogastric tubes, indwelling endotracheal tubes, or tracheostomy tubes [9–12]. In the current era of imaging, patients are often referred when a vascular ring is discovered, but the patient appears to be "asymptomatic." Careful questioning of these patients often reveals symptoms related to their respiratory function and swallowing.

Double Aortic Arch

Infants with a double aortic arch tend to present earlier in life than patients with a right aortic arch. The great majority of these patients will have symptoms before 1 month of age. This is in contrast to patients with a right aortic arch where nearly 50 % do not have symptoms until after 1 month of age. There are three main anatomic types of double aortic arch. The most common is a dominant right aortic arch with a smaller left aortic arch (80 %) (Fig. 123.8). A dominant left

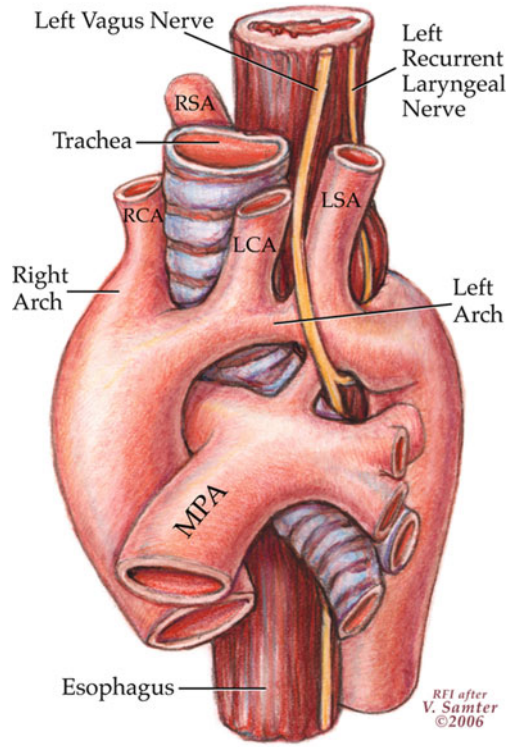

Fig. 123.8 Double aortic arch. Right (posterior) arch is dominant. The smaller left arch is patent. *LCA* left carotid artery, *LSA* left subclavian artery, *MPA* main pulmonary artery, *RCA* right carotid artery, *RSA* right subclavian artery (Reprinted with permission from: Backer CL, Mavroudis C, Stewart RD, Holinger LD. Congenital anomalies: vascular rings. In: Patterson GA, Cooper JD, Deslauriers J, Lerut AE, Luketich JD, Rice TW (eds): Pearson's Thoracic and Esophageal Surgery, Philadelphia, Church Livingstone Elsevier, 2008, 242–255)

aortic arch is found in 10 % of patients. "Balanced" aortic arches are found in the remaining patients (10 %) although these patients often have subtle signs that one arch is larger than the other, on CT imaging (Fig. 123.2). The most common approach to the double aortic arch is through a left thoracotomy using a muscle-sparing technique. However, for the patient with a dominant left aortic arch, an approach through a right thoracotomy is recommended. This emphasizes the importance of precise preoperative imaging using the CT scan. The chest is entered through the fourth intercostal space. The pleura overlying the vascular ring is opened. This opening in the pleura should be somewhat anterior rather than

posterior in order to avoid the thoracic duct. Careful dissection is performed to clearly identify all of the pertinent vascular structures. In particular, the recurrent laryngeal nerve must be carefully identified and carefully protected from injury. Some patients with double aortic arch will have atresia of the smaller arch at the distal portion of the arch. This occurs in approximately one-third of patients with a double aortic arch. The site of atresia is usually where the lesser arch inserts into the descending thoracic aorta. There are occasional patients who will have coarctation of one or both of the aortic arches.

The surgical intervention should be directed at dividing the smaller of the two arches at a site that does not compromise the flow of blood to the head vessels or lower body. The intended arch to be divided is temporarily occluded while the anesthesiologist carefully checks blood pressure and pulses above and below the site of the temporary occlusion. The use of pulse oximetry can facilitate this portion of the procedure. The arch is divided between vascular clamps, and the divided stumps are oversewn with polypropylene suture (Fig. 123.9). These authors recommend dividing only a portion of the arch and placing sutures on each side of the partially divided arch before completely dividing the arch. This helps ensure the arch does not slip out of the vascular clamp. The Potts ductus clamp which has small teeth to grip the arch is used in our practice. The oversewn arch should be reinforced with several interrupted prolene sutures. This helps to ensure prevention of catastrophic hemorrhage. When the stumps divide, they typically separate by 1–2 cm (Fig. 123.10). They may also disappear into the posterior mediastinum. Hence, precise hemostasis prior to clamp release is of paramount importance.

Some surgeons have reported thoracoscopic division of vascular rings using hemoclips [11–13]. This technique is not used at our institution. There is a risk of hemorrhage should a clip slip off a vessel when the vessel retracts. Hence, this technique is most applicable for those patients who have an atretic arch. However, our experience using a muscle-sparing thoracotomy has been that there is limited morbidity from the incision. The advantage of full exposure and

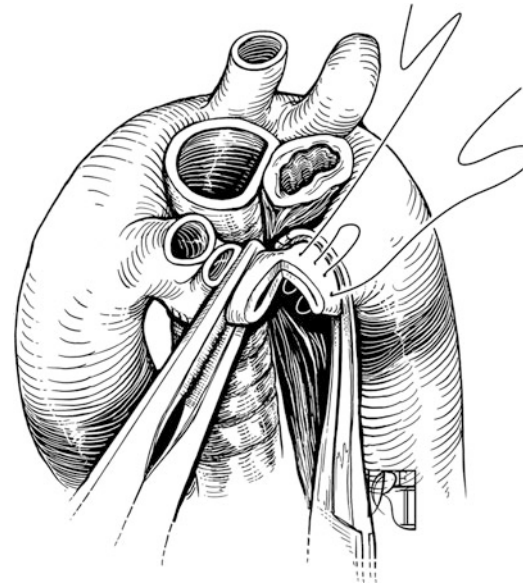

Fig. 123.9 Surgical division of a double aortic arch. Through a left thoracotomy, the smaller left aortic arch has been occluded with vascular clamps. Staged division and s demonstrated (Reprinted with permission: Backer CL, Mavroudis C. Vascular Rings and Pulmonary Artery Sling. In: Mavroudis C, Backer CL (eds) Pediatric Cardiac Surgery, 4th ed., Oxford, UK, pp. 234–255)

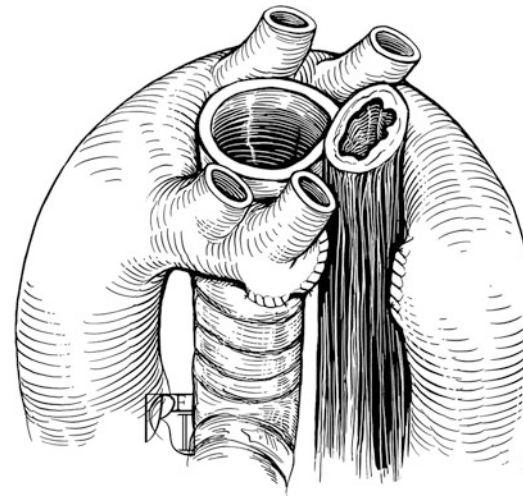

Fig. 123.10 Completed division and oversewing of left aortic arch and left ligamentum. The compression on the esophagus and trachea is relieved (Reprinted with permission: Backer CL, Mavroudis C. Vascular Rings and Pulmonary Artery Sling. In: Mavroudis C, Backer CL (eds) Pediatric Cardiac Surgery, 4th ed., Oxford, UK, pp. 234–255)

access to vessels should there be any bleeding outweighs the cosmetic advantage of the thoracoscopic approach.

At the conclusion of the operation, the mediastinal pleura is left widely open. This helps decrease the chance that recurrent scar tissue could form and cause recurrent stenosis in the area of the ring division. In these authors' practice, it has been noted that in several patients operated elsewhere who have had recurrence of their symptoms and at reoperation, it was noted that the pleura was closed at the initial operation. These patients developed significant scar tissue which was quite dense.

The operative repair is completed by dividing any adhesive bands surrounding the esophagus in the area of the divided vascular ring. Division of these small bands was emphasized by Gross in his earlier papers [1]. In the current era, all patients are drained with a small Silastic Blake drain (Ethicon, Inc., Somerville, NJ). These drains are very soft and well tolerated. They help ensure rapid diagnosis should there be any hemorrhage in the postoperative period and are also useful for demonstrating chylothorax prior to significant pleural effusion accumulation. The child is extubated in the operating room and monitored in the cardiac care unit for 48–72 h. It should be noted that many patients have residual tracheal and/or bronchomalacia following the operation. The parents are counseled that it may take up to 1 year for the child's noisy breathing to disappear as this malacia caused by the vascular ring resolves over a period of time.

Results

Since the first successful case performed at Ann & Robert H. Lurie Children's Hospital of Chicago in 1947 by Dr. Willis Potts, 145 patients have had repair of a double aortic arch. Mean age was 2.86 years and median age 1.17 years. There has been no operative mortality since 1952. Two patients have required a reoperation for recurrent symptoms. In both cases, this occurred from scar tissue that developed at the site of the divided posterior left arch. There has been a 2 % incidence of chylothorax. Several of these patients have been treated by rapid return to the operating room (3–5 days) for oversewing of a leaking lymphatic at the site of the dissection. This has improved the postoperative length of stay following this complication. In other patients, rapid resolution of the chylothorax has occurred with the adoption of a fat-free diet. This management has been facilitated by the use of the Blake drain at the time of the operation for monitoring the pleural fluid character.

Right Aortic Arch

These children typically present somewhat later in life than patients with a double aortic arch. Most of these patients develop symptoms between 1 and 6 months of age. The ring is anatomically "looser" as it is partially formed by the low-pressure pulmonary artery and the ligamentum which does not have blood flow. There are two primary branching patterns of the brachiocephalic vessels in patients with an aortic arch. The first is a retroesophageal left subclavian artery (Fig. 123.11) and the second is mirror-image branching (Fig. 123.12). Sixty-five percent of the patients have a retroesophageal left subclavian artery, and 34 % of the patients have mirror-image branching. With a retroesophageal left subclavian artery, the first branch off of the ascending aorta is the left carotid artery, the second branch is the right carotid artery, and the third branch is the right subclavian artery. The left subclavian artery originates from the descending thoracic aorta adjacent to the takeoff of the ligamentum arteriosum. It is the base of this left subclavian artery that is frequently formed by a dilatation referred to as "Kommerell diverticulum" [14]. The Kommerell diverticulum is a remnant of the fourth aortic arch. In patients with mirror-image branching, the first branch off of the ascending aorta is a left innominate artery that contains both the left carotid and the left subclavian arteries. The second branch is the right carotid artery. The third branch is the right subclavian artery. In these patients, the ligamentum

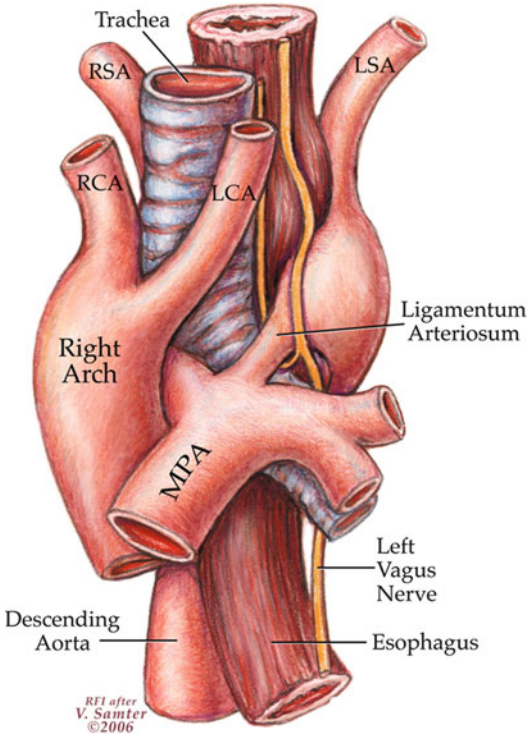

Fig. 123.11 Right aortic arch, retroesophageal left subclavian artery, and left ligamentum arteriosum. *LCA* left carotid artery, *LSA*, left subclavian artery, *MPA* main pulmonary artery, *RCA* right carotid artery, *RSA* right subclavian artery (Reprinted with permission. Backer CL, Mavroudis C, Stewart RD, Holinger LD. Congenital anomalies: vascular rings. In: Patterson GA, Cooper JD, Deslauriers J, Lerut AE, Luketich JD, Rice TW (eds): Pearson's Thoracic and Esophageal Surgery, Philadelphia, Church Livingstone Elsevier, 2008, 242–255)

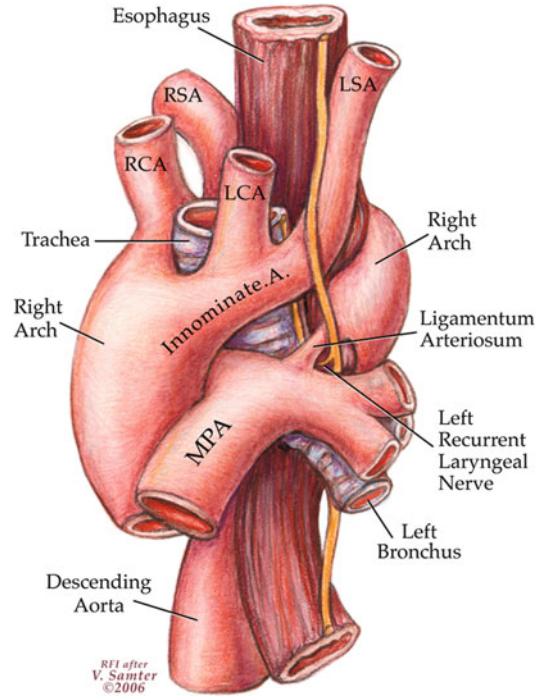

Fig. 123.12 Right aortic arch, mirror-image branching. In this case a vascular ring is formed because the ligamentum inserts into the descending aorta. *Innominate A* innominate artery, *LCA* left carotid artery, *LSA* left subclavian artery, *MPA* main pulmonary artery, *RCA* right carotid artery, *RSA* right subclavian artery (Reprinted with permission. Backer CL, Mavroudis C, Stewart RD, Holinger LD. Congenital anomalies: vascular rings. In: Patterson GA, Cooper JD, Deslauriers J, Lerut AE, Luketich JD, Rice TW (eds): Pearson's Thoracic and Esophageal Surgery, Philadelphia, Church Livingstone Elsevier, 2008, 242–255)

arteriosum either may insert into the descending thoracic aorta causing a vascular ring (Fig. 123.13) or may originate from the innominate artery, not causing a vascular ring (Fig. 123.14a, b). Right aortic arch is much more common in patients with tetralogy of Fallot and in patients with common arterial trunk. However, these patients do not appear to have an increased incidence of vascular rings because in many of these cases, the ligamentum originates from the innominate artery.

For patients with a right aortic arch and left ligamentum, the surgical approach is through a muscle-sparing left thoracotomy, again through the fourth intercostal space. The lung is retracted anteriorly and the pleura is opened. This incision should be relatively close to the vagus nerve, but not so close as to injure the vagus and left recurrent laryngeal nerve. Again, this will help avoid entry into the thoracic duct which is typically found in the more posterior mediastinum. The ligamentum is carefully dissected. The ligamentum can be divided either between silk ligatures reinforced with prolene suture or divided between vascular clamps with oversewing of the two stumps (Fig. 123.15a–c). Release of the ligamentum should lead to a separation of the two stumps of the ligamentum by 1–2 cm.

There is a subset of patients as mentioned earlier who have an origin of the left subclavian

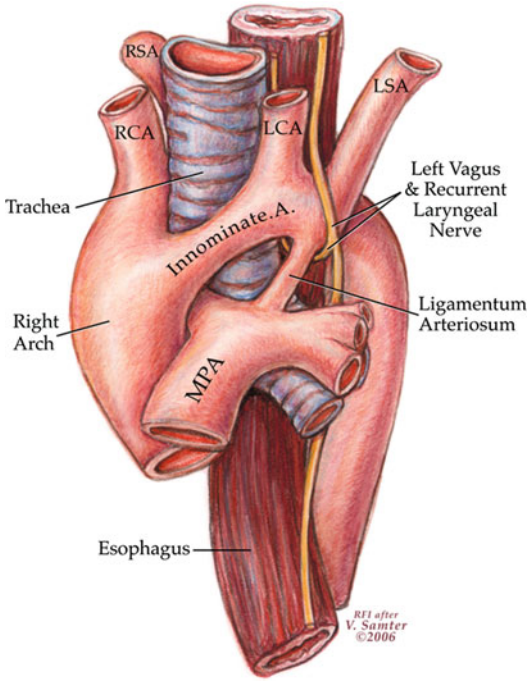

Fig. 123.13 Right aortic arch, mirror-image branching with ligamentum from innominate artery. In this case, a vascular ring is not formed. *Innominate A* innominate artery, *LCA* left carotid artery, *LSA* left subclavian artery, *MPA* main pulmonary artery, *RCA* right carotid artery, *RSA* right subclavian artery (Reprinted with permission. Backer CL, Mavroudis C, Stewart RD, Holinger LD. Congenital anomalies: vascular rings. In: Patterson GA, Cooper JD, Deslauriers J, Lerut AE, Luketich JD, Rice TW (eds): Pearson's Thoracic and Esophageal Surgery, Philadelphia, Church Livingstone Elsevier, 2008, 242–255)

artery from a Kommerell diverticulum. A significant Kommerell diverticulum is defined as an aneurysmal bulging of the base of the subclavian artery which is more than 1½ to 2 times the size of the subclavian artery. In many of these patients, the Kommerell diverticulum is an independent cause of the compression of the posterior portion of the esophagus and trachea. This will be visible on the CTA (Fig. 123.5a, b) and on the preoperative bronchoscopy. Should there be a significant Kommerell diverticulum, resection of the latter and transfer of the left subclavian artery to the left carotid artery is recommended (Fig. 123.15a–c). This operation is clearly more complex than simple ligamentum division. The patient is given 100 units/kg of heparin prior to applying the vascular clamps. The base of the Kommerell diverticulum is occluded with a vascular clamp that also occludes a portion of the wall of the descending thoracic aorta (Fig. 123.15a). Flow to the lower body is maintained by assessing the lower extremity blood pressure with a test occlusion of the clamp. The left subclavian artery can be controlled by a small vascular hemoclip. The Kommerell diverticulum is resected (Fig. 123.15b). The stump on the aorta is carefully oversewn with running prolene suture in two layers. This is reinforced with multiple interrupted prolene mattress sutures prior to releasing the clamp. Staged oversewing and division techniques are recommended here to prevent slipping of the

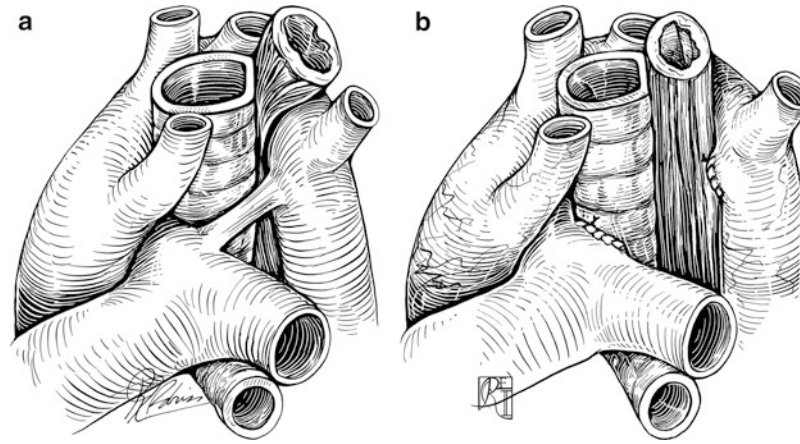

Fig. 123.14 Surgical division of left ligamentum in a patient with a right aortic arch. (**a**) Anatomy of vascular ring. (**b**) Post-division and oversewing of ligamentum (Reprinted with permission: Backer CL, Mavroudis C. Vascular Rings and Pulmonary Artery Sling. In: Mavroudis C, Backer CL (eds) Pediatric Cardiac Surgery, 4th ed., Oxford, UK, pp. 234–255)

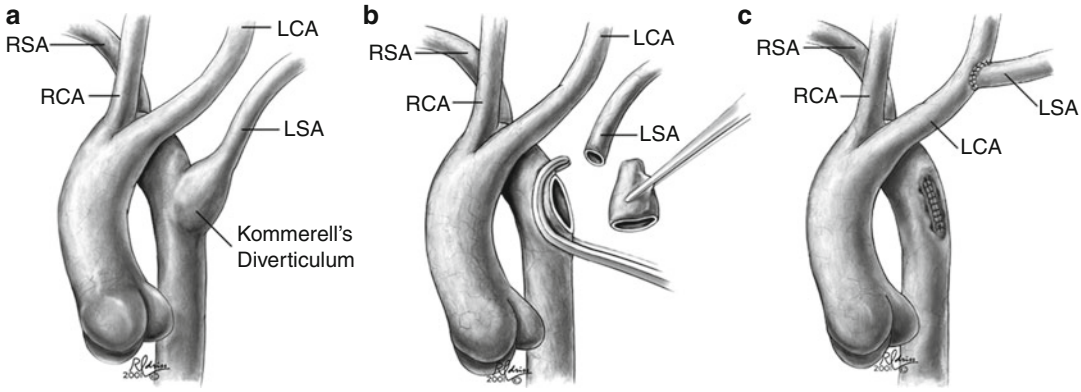

Fig. 123.15 (**a**) The anatomy of a patient with a right aortic arch, retroesophageal left subclavian artery, and large Kommerell diverticulum. The Kommerell diverticulum is an embryologic remnant of the left fourth aortic arch. *LCA*/*RCA* left/right carotid artery, *LSA*/*RSA* left/right subclavian artery. (**b**) Resection of a Kommerell diverticulum through a left thoracotomy. There is a vascular clamp partially occluding the descending thoracic aorta at the origin of the Kommerell diverticulum. The Kommerell diverticulum has been completely resected. The clamp on the distal left subclavian artery is not illustrated. (**c**) The completed repair. The orifice where the Kommerell diverticulum was resected is usually closed primarily, or, as shown in the inset, the orifice can be patched with polytetrafluoroethylene if necessary. The left subclavian artery has been implanted into the side of the left common carotid artery with fine running polypropylene suture. Resection of Kommerell's diverticulum and left subclavian artery: transfer for recurrent symptoms after vascular ring division (Reprinted with permission. Eur J Cardiothorac Surg. 2002;221:64–69)

vessels through the clamps. The left common carotid artery is then identified in the posterior mediastinum. Typically, it lies in a space where the vagus nerve is anterior and the left recurrent laryngeal nerve as it recurs up towards the neck where it is posterior. It is in a plane between these two structures that the common carotid artery can be identified. The artery is encircled in a vessel loop and is pulled into the field. A vascular clamp is placed to completely occlude the artery. Arteriotomy is performed and an anastomosis to the left subclavian artery is performed with running prolene suture. The system is de-aired and the clamps are released (Fig. 123.15c).

Results

Since 1947, 153 patients have had division of the ligamentum for a right aortic arch and left ligamentum (mean age of 4.09 years and median age of 1.52 years). There has been no operative mortality since 1959. Two patients in our series have required reoperation for later resection of a Kommerell diverticulum. Sixteen patients from other institutions have undergone a reoperation for Kommerell diverticulum with left subclavian artery transfer [15]. Since 2001, 20 patients have had primary excision of a Kommerell diverticulum; 15 of these patients had simultaneous division and reimplantation of the left subclavian artery into the left carotid artery [16]. The mean age at operation in this group was higher than in the patients with only ligamentum division. These patients were 9 ± 6 years of age. There were no complications related to the subclavian artery transfer, and no patient had a recurrent laryngeal nerve injury. Two patients had a postoperative chylothorax. In all patients, there was resolution of their preoperative airway and esophageal symptoms.

Pulmonary Artery Sling

Pulmonary artery sling was first reported in 1897 as an autopsy finding in a 7-month-old infant who died of respiratory distress [17]. The left pulmonary artery originates from the right pulmonary artery and encircles the distal trachea coursing between the trachea and esophagus to the hilum of the left

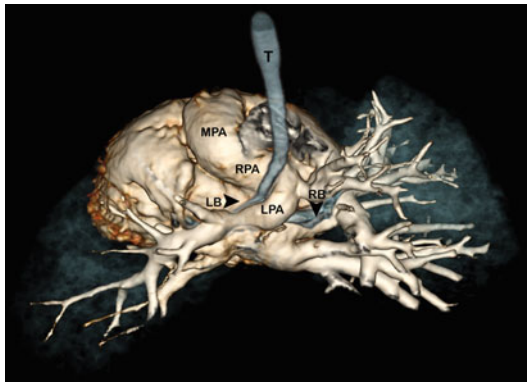

Fig. 123.16 Pulmonary artery sling. Contrast-enhanced chest CT angiography 3D multiplanar reformat. Posterior view of an anomalous origin of the left pulmonary artery from the right pulmonary artery with aberrant course posterior to the trachea. There is distal tracheal stenosis secondary to complete tracheal rings and compression of the origin of the right mainstem bronchus. *LB* left bronchus, *LPA* left pulmonary artery, *MPA* main pulmonary artery, *RB* right bronchus, *RPA* right pulmonary artery, *T* trachea

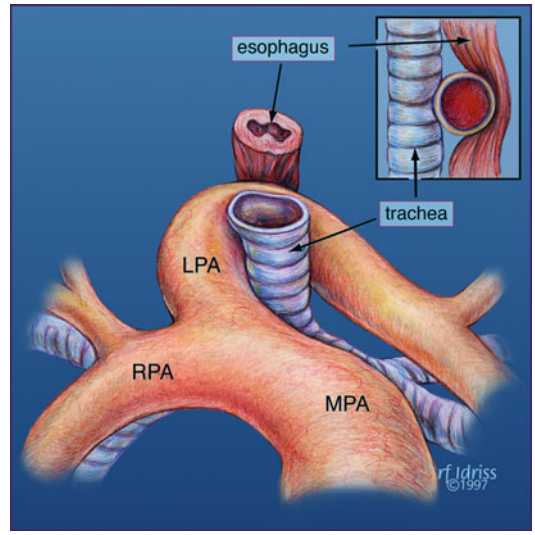

Fig. 123.17 Illustration of pulmonary artery sling (*anterior view*). The left pulmonary artery courses between the trachea and esophagus causing anterior compression of the esophagus. *LPA* left pulmonary artery, *MPA* main pulmonary artery, *RPA* right pulmonary artery

lung (Fig. 123.16). This left pulmonary artery acts as a sling applying pressure to both the right main bronchus and the lower portion of the trachea. In our series, there were several critically ill patients who presented with respiratory distress and required intubation. These patients were then diagnosed to have a pulmonary artery sling by echocardiography at the bedside. Other patients have presented with tracheal stenosis and the ability of the referring institution to only intubate the patient with a very small endotracheal tube. In our series of patients with pulmonary artery sling, 75 % have had tracheal stenosis secondary to complete cartilaginous tracheal rings. All patients with a diagnosis of pulmonary artery sling have had preoperative airway imaging with rigid bronchoscopy. Since the year 2000, all patients with pulmonary artery sling have had computed tomographic imaging of the chest with 3-dimensional reconstruction. Patients with pulmonary artery sling and complete tracheal rings are referred to as having the "ring-sling complex" [8].

The surgical repair of pulmonary artery sling should be undertaken as soon as the diagnosis is made because of the tenuous nature of the respiratory status in these children. One child in our series presented with an apneic episode with swallowing. The median age at surgery in our series is 3 months. Since 1985, 34 patients undergoing pulmonary artery sling repair at our institution have had this operation performed through a median sternotomy using cardiopulmonary bypass and mild hypothermia (Fig. 123.17) [18]. Associated intracardiac anomalies were repaired in seven patients. There have been no early deaths or complications related to the use of cardiopulmonary bypass. Median hospital stay was 24 days. The trachea repair has included pericardial patch tracheoplasty (n = 7), tracheal autograft (n = 10), tracheal resection (n = 4), and slide tracheoplasty (n = 5). There were four patients who had a severely hypoplastic or absent right lung. The primary technique of repair was to transect the left pulmonary artery at its origin from the right pulmonary artery and pass it through the mediastinum posterior to the trachea. The left pulmonary artery is then anastomosed to the main pulmonary artery anterior to the trachea (Fig. 123.18). In the four patients with a hypoplastic right lung, the left pulmonary artery was simply translocated anterior to the trachea during the tracheal repair [19, 20].

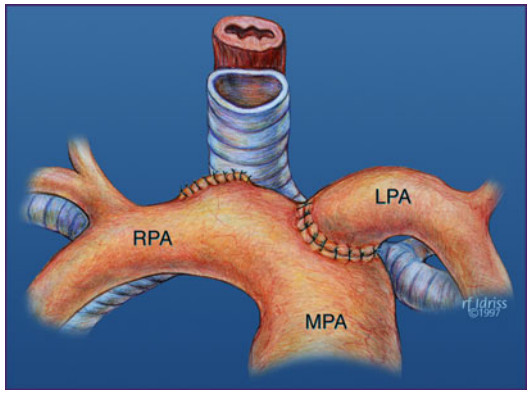

Fig. 123.18 Repaired pulmonary artery sling. The original origin of the left pulmonary artery has been oversewn. The left pulmonary artery has been reimplanted into the main pulmonary artery anterior to the trachea. *LPA* left pulmonary artery, *MPA* main pulmonary artery, *RPA* right pulmonary artery

The patency of the left pulmonary artery using this technique is 100 % with the mean percent blood flow to the left lung by nuclear scan being 41 % [18]. There were four late deaths in this series. Two were results of complications of tracheal surgery, one 6 months following pericardial patch tracheoplasty and the other nearly 2 years following graft tracheal surgery. One child died of biliary atresia 3 months after the operation, and one child died of pneumonia 6 years after the operation.

Complete Tracheal Rings

Our experience with the high frequency of tracheal stenosis in patients with pulmonary artery sling has led to a substantial experience in managing patients with tracheal stenosis secondary to complete cartilage tracheal rings. These patients frequently present with respiratory distress in infancy. Medical management of tracheal stenosis is associated with a 40 % mortality rate [21]. Patients are often referred when even the smallest endotracheal tube cannot be passed very far below the vocal cords. In all cases, the diagnosis is made by rigid bronchoscopy. In addition, CT is performed to find the extent of the tracheal stenosis. A total of 80 patients have now undergone repair of complete tracheal rings at our institution: pericardial tracheoplasty (n = 28), resection with end-to-end anastomosis (n = 14), slide tracheoplasty (n = 19), and free tracheal autograft (n = 19). Historically, the pericardial tracheoplasty was the first operation used to successfully treat patients with tracheal stenosis and was first performed by Farouk S. Idriss at our institution [22]. This was also the first use of cardiopulmonary bypass for tracheal repair in an infant.

Tracheal Stenosis Procedures

For the pericardial tracheoplasty, the patient is placed on cardiopulmonary bypass via median sternotomy incision. The aorta is retracted to the left. The anterior surface of the trachea is dissected. An incision is made through the anterior wall of the trachea throughout the length of the tracheal stenosis. An autologous pericardial patch is then anchored in place with interrupted absorbable sutures such as PDS or Vicryl (Fig. 123.19a, b). The pericardium is tacked to adjacent mediastinal structures to prevent tracheomalacia. Although our operative mortality with this technique was low, there was significant late mortality with a total mortality rate of 18 %. This was due to patch tracheomalacia, granulation tissue, and the need for a tracheostomy [23]. Other surgeons have reported excellent results with pericardial tracheoplasty [24].

In 1996, a technique called the tracheal autograft operation was introduced into practice at Lurie Children's [25]. In this procedure, the patient is again approached through a median sternotomy using cardiopulmonary bypass. The trachea is incised throughout the length of the tracheal stenosis. The midportion of the tracheal stenosis is resected, usually six to seven cartilage rings, and this piece is then used as a free tracheal autograft anteriorly. The two opened portions of the trachea are brought together posteriorly end-to-end. The autograft then either fills the space anteriorly or is augmented with a small piece of pericardium. This technique was performed in 20 patients, but the long-term mortality rate was 27 %. This was related to problems with

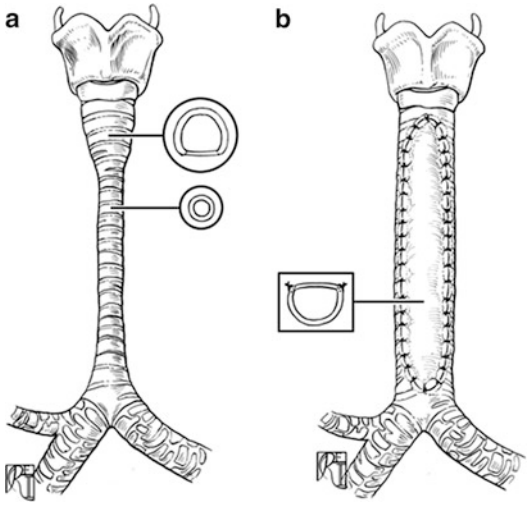

Fig. 123.19 Pericardial patch tracheoplasty technique. (**a**) Long-segment tracheal stenosis with complete tracheal rings. Note the absence of the membranous trachea. The trachea is incised anteriorly through the extent of the complete tracheal rings. (**b**) Completed repair with trachea patched anteriorly with autologous pericardial patch

granulation tissue, dehiscence in one case, and erosion of the posterior anastomosis into the carotid artery.

Tracheal resection has been performed with median sternotomy and cardiopulmonary bypass. The trachea is dissected and the area of the tracheal stenosis is carefully identified with flexible intraoperative bronchoscopy. The narrowed segment is sharply resected and a direct end-to-end anastomosis is performed with interrupted PDS suture. In this experience, there has only been one mortality using this technique in a very small infant. However, this approach is only applicable to patients with a short distance of tracheal stenosis, typically less than 1/3 of the trachea [26].

Since 2001, the slide tracheoplasty has been adopted as these authors' procedure of choice [27]. This operation has now been performed in 19 patients. There have been two deaths, one in a patient who had associated pulmonary atresia and the other in a patient with agenesis of the right lung. The slide tracheoplasty is performed with median sternotomy and cardiopulmonary bypass. The midportion of the tracheal stenosis is carefully evaluated with intraoperative flexible fiber-optic bronchoscopy. The trachea is transected in the midportion of the stenosis (Fig. 123.20a, b). Two incisions are then made in the upper and lower portions of the remaining trachea. The lower trachea is incised anteriorly and the upper trachea is incised posteriorly. These incisions can be reversed if the patient had a prior tracheostomy that needs to be encompassed in the repair. We then slide the two ends together and perform a running anastomosis with PDS suture (Fig. 123.20b, c). This is now our procedure of choice for patients with long-segment tracheal stenosis. These patients are extubated 1–5 days after the operation. They all have bronchoscopic evaluation prior to discharge from the hospital. The largest reported series are from Great Ormond Street and Cincinnati Children's Hospital. The authors from Great Ormond Street reported 84 patients with a mortality rate of 13 % [28]. Manning reported 80 patients with a 5 % mortality [29].

Innominate Artery Compression Syndrome

A small number of patients will have significant compression of the anterior portion of the trachea by the innominate artery as it crosses from left to right after originating from the ascending thoracic aorta. This is not a vascular ring per se, but an external tracheal compression. It is unclear why in some patients this causes significant compression of the trachea. In the authors' institution, the indications for operating on these patients are symptoms such as apnea or cyanotic spells and bronchoscopic evidence of compression of more than 80 % of the tracheal lumen. CT imaging is also used to make this diagnosis. This operation was originally described by Gross using a left thoracotomy at Boston Children's Hospital [30]. At our institution, we have used a small right submammary thoracotomy for this operation. The right lobe of the thymus is excised in order to identify the innominate artery. Excising the thymus may contribute to improving the room in the

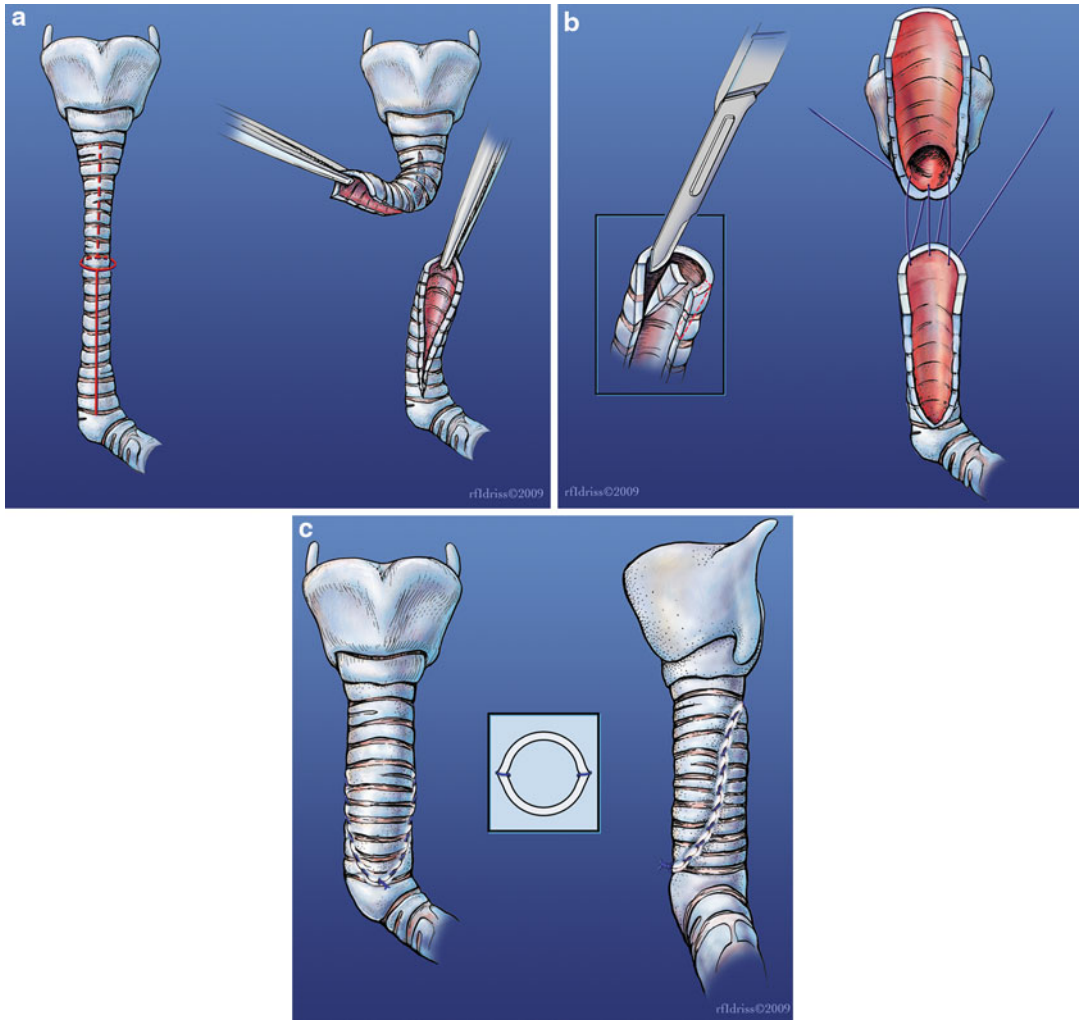

Fig. 123.20 Slide tracheoplasty; absent right lung. (**a**) The patient has been placed on cardiopulmonary bypass with mild hypothermia to 32 °C. The trachea is transected in the midportion of the tracheal stenosis. This site is determined either by external examination or by internal bronchoscopic findings. The inferior portion of the trachea is incised anteriorly, and the superior portion of the trachea is incised posteriorly. (**b**) The ends of the trachea are beveled as shown in the small inset. The anastomosis is performed with running 6.0 polydioxanone suture. The suture line is started superiorly (parachute technique) and finished inferiorly just above the carina. (**c**) Completed slide tracheoplasty. The everting running suture line helps to avoid the "figure of 8" configuration problem after the completed repair

mediastinum for the innominate artery. The innominate artery is then suspended to the posterior table of the sternum with three interrupted pledgeted sutures (Fig. 123.21). Confirmation of the results of the suspension is obtained by intraoperative bronchoscopy. The right radial pulse must be carefully monitored either with an arterial line, pulse oximetry, or blood pressure cuff to make sure that this vessel is not compromised. Currently, we have only been performing this operation on approximately one patient per year. Another operation that has been used at some institutions is to transect the innominate artery through a median

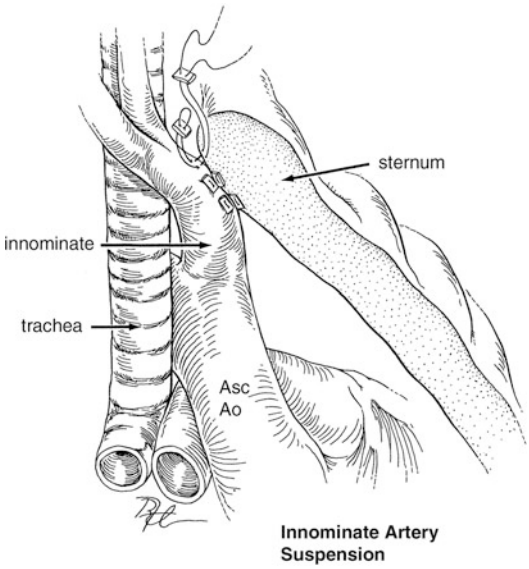

Fig. 123.21 Innominate artery suspension. The innominate artery is suspended to the posterior sternal table with interrupted pledgeted sutures. *Asc Ao*, ascending aorta (Reprinted with permission. Backer CL, Mavroudis C. Vascular Rings and Pulmonary Artery Sling. In: Mavroudis C, Backer CL (eds) Pediatric Cardiac Surgery, 4th ed., Oxford, UK, pp. 234–255)

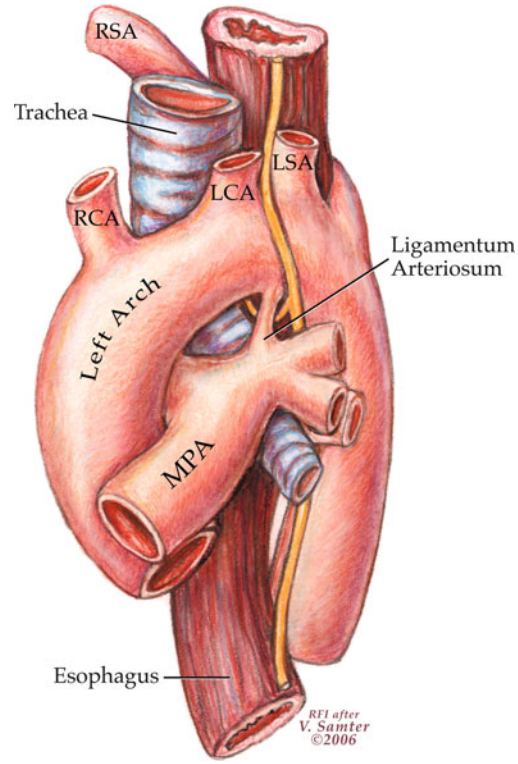

Fig. 123.22 Left aortic arch with aberrant right subclavian artery. *LCA* left carotid artery, *LSA* left subclavian artery, *MPA* main pulmonary artery, *RCA* right carotid artery, *RSA* right subclavian artery

sternotomy incision and reimplant it into the ascending aorta at a site more rightward and anterior so the trachea is not compressed [31]. We have not had experience with that procedure. In our series, only two patients have required reoperation for resuspension.

Aberrant Right Subclavian Artery

One of the most common vascular anomalies in humans is origin of the right subclavian artery from the descending thoracic aorta (Fig. 123.22). This occurs in 0.5 % of all humans [32]. Because it is so common in patients who have dysphagia, it has been blamed in the past for the swallowing symptoms. This has earned this anatomic malformation the label "dysphagia lusoria." In most cases, the aberrant right subclavian artery is actually a red herring and not the true etiology of the child's symptoms [33]. We have not operated on a child with this diagnosis since 1973.

Rare Vascular Rings

As mentioned in the "Embryology" section, there are many possible configurations of malformations depending on which portions of the aortic arch are deleted or remain present. One unusual combination is that of a left aortic arch with right-sided descending thoracic aorta. If there is a right patent ductus arteriosus or ligamentum arteriosum, a vascular ring is formed [34]. In most of these patients, a left aortic arch is actually a cervical arch resulting from persistence to the third rather than the fourth embryonic arch [35]. The cervical arch may independently compress the trachea in the absence of a right ductus. This is one of the few vascular rings best approached through a right thoracotomy rather than a left thoracotomy [36]. In the old literature, there are cues from the

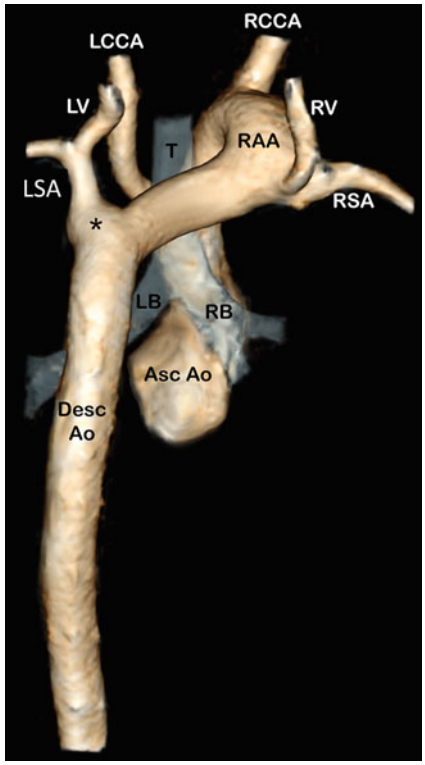

Fig. 123.23 3D MPR posterior view shows circumflex aorta in a 5-year-old child with a right cervical aortic arch, left ligamentum arteriosum (*), and retroesophageal left subclavian artery. The descending thoracic aorta crosses from right to left posterior to the trachea and superior to the carina. The posterior compression of the trachea is not relieved by ligamentum division. *Asc Ao* ascending aorta, *Desc Ao* descending aorta, *LB* left bronchus, *LCCA* left common carotid artery, *LSA* left subclavian artery, *LV* left vertebral, *RAA* right aortic arch, *RB* right bronchus, *RCCA* right common carotid artery, *RSA* right subclavian artery, *RV* right vertebral, *T* trachea

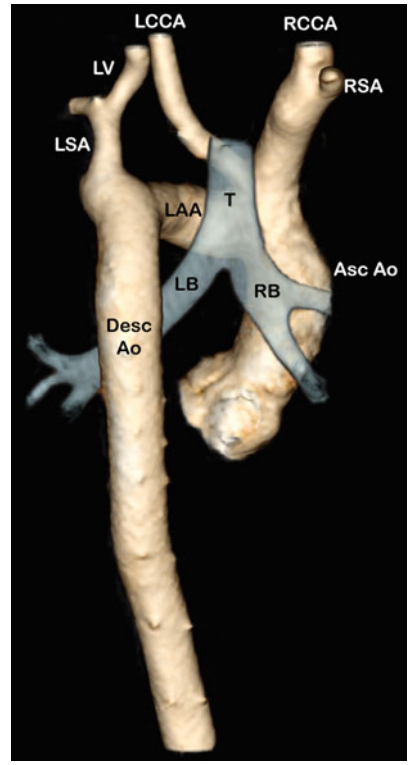

Fig. 123.24 Postoperative CTA 3D MPR posterior view shows circumflex aorta (as shown in Fig. 123.23) in a 5-year-old child. The transverse aortic arch is now anterior and to the left of the trachea. The anastomosis of the descending thoracic aorta to the transverse arch in this patient was widely patent and is off to the left of the trachea not compressing the trachea. *Asc Ao* ascending aorta, *Desc Ao* descending aorta, *LB* left bronchus, *LCCA* left common carotid artery, *LSA* left subclavian artery, *LV* left vertebral, *RAA* right aortic arch, *RB* right bronchus, *RCCA* right common carotid artery, *RSA* right subclavian artery, *RV* right vertebral, *T* trachea

chest x-ray that helped lead to this diagnosis. In the current era with computed tomographic imaging of all patients, this can be carefully planned prior to the operation, and the approach through the right thoracotomy will become more apparent. We reported a rare patient with a right aortic arch and a right ligamentum [37].

There is another group of patients with a rare vascular ring called a circumflex aorta. These patients have a right aortic arch, but the descending thoracic aorta traverses from right to left superior to the carina and independently compresses the trachea posteriorly (Fig. 123.23). Repair of this is done by an aortic uncrossing operation [38]. This operation is performed through a median sternotomy with cardiopulmonary bypass, deep hypothermia, and circulatory arrest. The aorta is transected on the right side just distal to the takeoff of the right subclavian artery. This portion of the aorta is then oversewn. The descending thoracic aorta is mobilized posterior to the esophagus and brought up anterior to the trachea and esophagus and then anastomosed to the side of the ascending aorta (Fig. 123.24). This completely relieves the posterior

compression. We have now performed this operation successfully in four patients; the mean age in that series was 3 years of age [39].

Conclusion

The diagnosis of vascular ring or pulmonary artery sling should be suspected in any infant or child presenting with symptoms of respiratory distress, noisy breathing, or dysphagia. Suspicion of the diagnosis is often generated by the plain chest x-ray. However, the diagnosis is best established by CT imaging which accurately delineates the anatomy of the vascular ring and the associated tracheal pathology. All patients diagnosed with a vascular ring should have an echocardiogram because of the 12 % incidence of associated congenital heart disease. Bronchoscopy should be performed in all cases to assess for additional tracheal pathology and to provide a clinical assessment of the degree of tracheomalacia and bronchomalacia. The common vascular rings are double aortic arch, right aortic arch with left ligamentum, and pulmonary artery sling. The surgical approach to double aortic arch is usually through a left thoracotomy although patients with a dominant left arch should be approached through a right thoracotomy. Patients with a right aortic arch and left ligamentum are approached through a left thoracotomy. Patients with double aortic arch and right aortic arch are treated by dividing the smaller of the two arches and/or the ligamentum. Patients with pulmonary artery sling are treated by median sternotomy with cardiopulmonary bypass for reimplantation of the left pulmonary artery anterior to the trachea and slide tracheoplasty if complete tracheal rings are associated. Innominate artery compression has been treated with a right thoracotomy and suspension of the innominate artery to the sternum. Close cooperation between the cardiothoracic and otolaryngology service is required to provide optimal care of these patients. There has been no operative mortality from an isolated vascular ring or pulmonary artery sling at Ann & Robert H. Lurie Children's Hospital of Chicago since 1959. Infants undergoing simple vascular ring repair have a 92 % incidence of freedom from respiratory symptoms at 1 year after the operation.

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