Vascular Rings and Pulmonary Sling

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7.1 Introduction

Vascular rings are a group of congenital anomalies, in which the aortic arch and its branches form a complete or partial ring around the trachea, the bronchi, and the esophagus, with potential extrinsic compression leading to variable degrees of respiratory problems or feeding difficulties.

They constitute 1 % of all the congenital vascular abnormalities [1], and the term vascular rings was originally introduced for the first time in 1945 by Gross, when he performed the first surgical division of a double aortic arch [2].

Pulmonary sling describes a congenital vascular anomaly involving the development of the pulmonary arteries, more often the left one. Usually, the anomalous left pulmonary artery arises from the posterior aspect of the right pulmonary artery; it courses over the right bronchus and running from right to left, posterior to the trachea or carina and anterior to the esophagus; it reaches the hilum of the left lung, forming a sling around the trachea.

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The unifying characteristic of all these anomalies is the presence of a complete or incomplete vascular structure (in some cases with fibrous replacement) surrounding the tracheoesophageal unit.

The clinical picture can range from asymptomatic patients to patients presenting with breathing disorders and/or swallowing difficulties due to the bronchial and tracheal and/or esophageal constriction [3]. Vascular rings and pulmonary slings may be associated with genetic defects, such as 22q11.2 deletion (DiGeorge syndrome), or they may be combined with cardiac conotruncal anomalies, as tetralogy of Fallot (ToF), double outlet right ventricle (DORV), pulmonary atresia (PA), truncus arteriosus (TA), and interrupted aortic arch (IAA) [4].

7.2 Embryology

During fetal development, the brachial vascular system, which will give rise to the aortic arch and its branches, develops and completes within the second and seventh gestational week.

The complex and dynamic embryological development consists of six paired aortic arches (although never present at the same time during fetal life) connecting the dorsal and the ventral aorta, which will reorganize with differential growth and reabsorption during the fetal period by processes of apoptosis and remodeling (Fig. 7.1).

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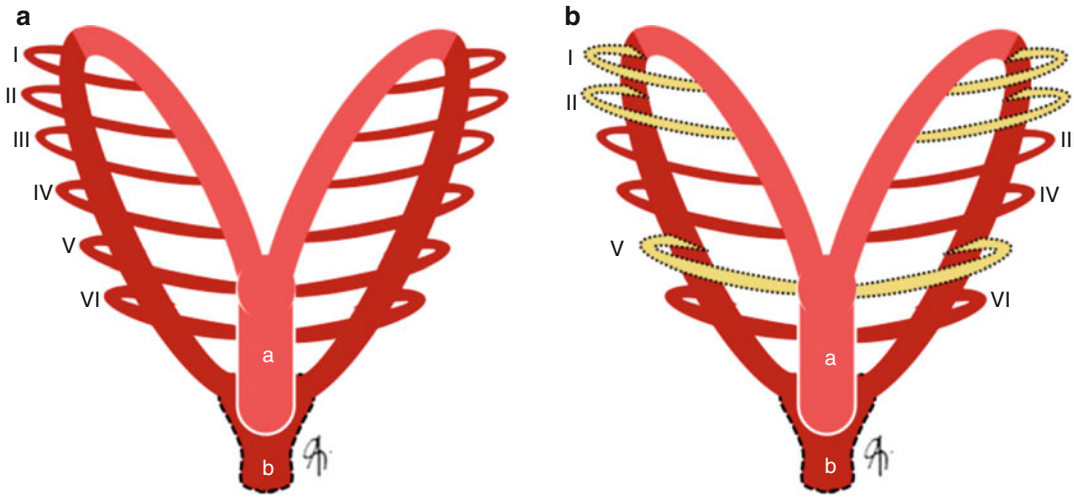

Fig. 7.1 The Rathke diagram: schematic representation of the six paired brachial arches (I–VI). (**a**) The brachial arches connect paired ventral aorta (*a*) and dorsal aorta (*b*). (**b**) Reabsorption of the first, second, and fifth arches

The first and second arches largely resorb and contribute to minor facial arteries; the fifth arches obliterate after the growth of the sixth ones; the third, along with the ventral aortic portion, will contribute to the development of the vessels arising from the mature aortic arch. The fourth will form the ultimate aortic arch, while the sixth will contribute to create the pulmonary arteries and the ductus arteriosus. On the right side, the segment of the dorsal aorta comprised between the fourth and sixth brachial arches will disappear; in the left side, it persists as the ductus arteriosus.

An unusual progression of obliterations and growth of this primitive vessel arrangement will result into a broad range of anatomical variations of vascular rings and slings [1].

A clear description of the possible anatomical variety is offered by Edward's scheme (Fig. 7.2); it condenses the conclusive phase of the brachial branches' embryogenesis, depicting the totipotent double (symmetrical) aortic arch, whose total or partial reabsorption or persistence may determine different anatomical variations.

In case of persistence of both left and right fourth brachial branches, a double aortic arch will result; if an obliteration of the portion of the right aortic arch between the subclavian artery and dorsal aorta occurs, a normal formation of a left aortic arch will take place; instead, if the reabsorption involves the opposite fourth arch, a right-sided aortic arch with so-called "mirror

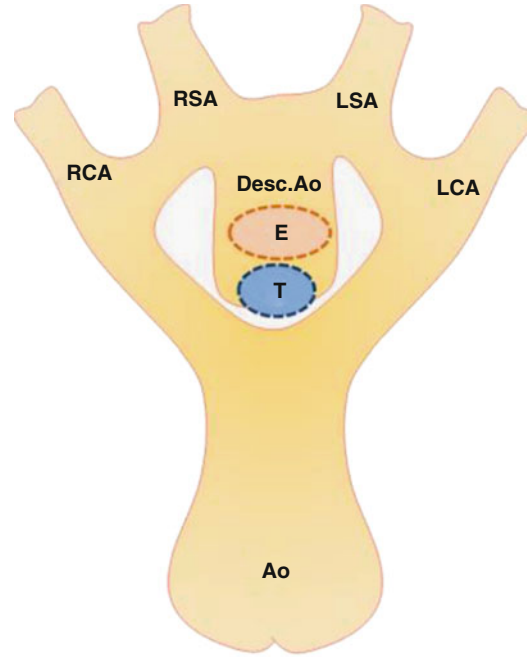

Fig. 7.2 Edward's diagram: totipotent double aortic arch system. Legend: *Ao* aorta; *Desc.Ao* descending aorta; *E* esophagus, *T* trachea, *RCA* right carotid artery, *RSA* right subclavian artery, *LCA* left carotid artery, *LSA* left subclavian artery

image" arrangement of arch vessel will result. Obliterations affecting other segments will induce, in addition to the left or right location of the arch, the presence of an aberrant subclavian artery (Fig. 7.3).

7.3 Classification

There are different classifications of vascular rings, some of which take into account the morphology of the ring and some others considering the position and anatomy of the arch.

In relation to the *morphology*, vascular rings can be divided into two large groups [5]:

1. *Complete*: when fibrovascular elements create a complete ring around the trachea and esophagus
2. *Incomplete*: if the trachea and esophagus are not completely encircled by fibrovascular structures

The most common abnormalities in the complete form group are *double aortic arch*, *right-sided aortic arch with left aberrant subclavian artery and Kommerell diverticulum*, and *right-sided aortic arch "mirror image"* type with ligamentum arteriosum on the left side.

Among the incomplete forms, the most frequent are *left aortic arch with right aberrant subclavian artery* and *pulmonary sling*.

Regarding the *position* of the aortic arch and branching pattern of the great vessels, different *anatomical* types can be identified:

1. Left aortic arch
   (a) Normal branching
   (b) Aberrant right subclavian artery
2. Right aortic arch
   (a) Mirror image
   (b) Aberrant left subclavian artery
3. Double aortic arch
4. Pulmonary artery sling
5. Cervical aortic arch

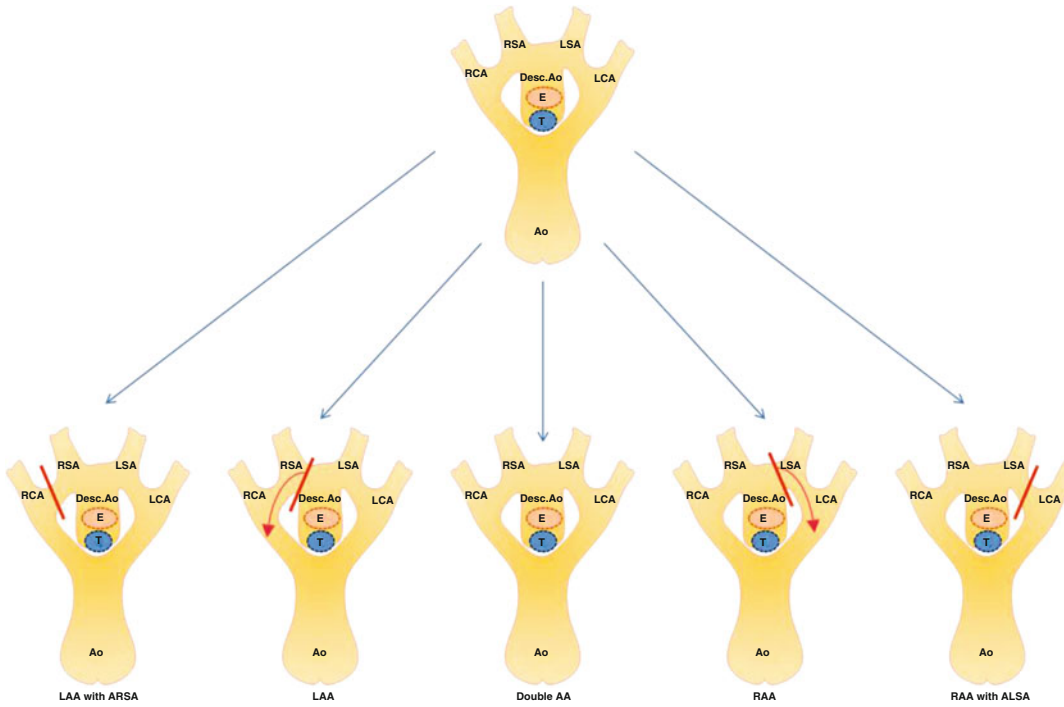

Fig. 7.3 Edward's diagram: hypothetical double arch system. Legend: *Ao* aorta, *Desc.Ao* descending aorta, *E* esophagus, *T* trachea, *RCA* right carotid artery, *RSA* right subclavian artery, *LCA* left carotid artery, *LSA* left subclavian artery, *LAA-ARSA* left aortic arch- aberrant right subclavian artery, *LAA* left aortic arch, *RAA* right aortic arch, *RAA-ALSA* right aortic arch-aberrant left subclavian artery, *Double AA* double aortic arch

Other anomalies exist, but they are either significantly less frequent or are usually asymptomatic.

7.4 Clinical Presentation and Diagnosis

Clinical presentation is heterogeneous (Table 7.1) and can vary from *asymptomatic* patients with incidental diagnosis during adulthood to *severe early symptoms* of either tracheal or esophageal compression, leading to early recognition of the more severe forms.

The breathing difficulty involves symptoms varying from wheezing, dry cough, and inspiratory dyspnea, with worsening during stress or meals. The wheezing can be misled as an asthmatic pathology.

Esophageal constriction is less frequent and characterized by increased risk of *ab-ingestis* events, gastroesophageal reflux, and progressive dysphagia, firstly at ingestion of solid food and only after for ingestion of liquids.

Eventually, recurrent infections of the respiratory tract can occur, in particular during the first year of patient life. Between 18 and 24 months, the symptoms tend to decline due to the physiological growth of body structures.

7.5 Imaging

Nowadays many possibilities exist to make a diagnosis in case of suspicion of vascular ring or pulmonary sling: *chest X-Ray (CXR), barium esophagram, echocardiography, computerized tomography scan* (*CT scan), magnetic resonance imaging (MRI), invasive angiography*, and *bronchoscopy.* All of these diagnostic techniques present advantages and disadvantages.

Table 7.1 Vascular ring and sling: clinical presentation

Asymptomatic	
Respiratory symptoms	Stridor, wheezing, chronic cough, recurrent respiratory infections, seal bark cough, tachypnea, intermittent cyanosis, asthma
Digestive symptoms	Feeding difficulty, dysphagia, recurrent emesis, gagging, choking, others

7.5.1 Chest X-Ray (CXR)

Chest radiography, with or without barium esophagography, can be considered the first-line imaging modality used in diagnosing tracheal or esophageal compression, particularly in children. CXR double projection (straight and leaning) can enlighten tracheal compression by nearby structures or tracheal displacement related to aortic arch location. In case of double aortic arch, the CXR image describes the trachea in axis, with two lateral compressions. The CXR with barium esophagography can display a posterior incision on the esophagus due to the presence of anomalous subclavian artery or Kommerell diverticulum, and in the presence of pulmonary sling, the cleft will be on the anterior wall of the esophagus. Moreover, there could be present signs of pulmonary atelectasis or lung hyperinflation in case of bronchial compression. Unfortunately, this technique does not allow direct images of vascular structures or vessels' anatomy, preventing from an accurate surgical planning.

7.5.2 Bronchoscopy

Bronchoscopy is nowadays an important tool to make a diagnosis when symptoms of respiratory distress are present. This procedure may demonstrate the presence of pulsatile compression and also the degree and precise location of such compression. In cases of pulmonary sling, bronchoscopy shows the length of hypoplastic tracheal segment, the diameter of this segment, and the presence of complete tracheal rings. Bronchoscopy is mandatory after surgical treatment to evaluate the results of surgery and in the postoperative period to follow the outcome of correction.

7.5.3 Echocardiography

The echocardiography can be considered the first tool to confirm the suspicion of vascular ring or sling. It is usually able to demonstrate the aortic side; the anomalies of vessels of the aortic arch, such as aberrant subclavian artery or aneurysm of the brachiocephalic trunk; the double aortic arch; and the anatomy of the pulmonary artery. However, echocardiography is a poor imaging tool to either establish or exclude the diagnosis of a vascular ring in case of poor acoustic windows such as in adult patients. In addition, it does not depict the ligamentous structures and hyperinflation of the lungs and the compression of the trachea or esophagus. Echocardiography is absolutely necessary for investigating associated cardiac defects.

7.5.4 Angiography

Angiography was the first procedure used to perform diagnosis in these patients until the last decade, but currently this imaging modality is rarely used, and it is useful only in selected cases. Angiography may be considered, if the anatomy of the patent vessels must be visualized, including the size of the two arches, in case of a double aortic arch, in order to make a proper selection of the arch that has to be cut. However, angiography is unreliable in demonstrating the tracheal or esophageal compression, and moreover it is an invasive procedure, requiring vascular access and the use of ionizing radiation and contrast agent.

7.5.5 Computed Tomography Scan (CT Scan)

Computed tomography angiography (CTA) allows for an accurate description of vascular and respiratory tract anatomy. Patent vascular channels are evident on CTA as contrast-enhancing segments and are well visualized on reconstructed 3D images. Conversely, atretic vascular segments and ligaments are not evident, but their presence can be inferred from traction on associated vascular structures or compression of the trachea [6]. Inspiratory and expiratory CTA studies allow the dynamic evaluation of tracheal caliber for narrowing or traction, which is particularly important in patients with associated tracheomalacia. CTA scanning times are shorter than magnetic resonance imaging (MRI), and therefore sedation is usually not necessary, which is a significant advantage in a young or respiratory distressed patient. The principal disadvantages of CTA are the need for intravenous contrast agents and the potential late consequences of radiation-dose exposure.

7.5.6 Magnetic Resonance Imaging (MRI)

MRI is currently the gold standard diagnostic modality, because of the anatomical definition of the anomaly with functional and dynamic study of the heart and vessels. Advantages of MRI over CTA include the freedom from exposure to both radiation and intravenous iodine contrast, as well as the ability to undertake functional studies in patients with intracardiac lesions. The limitations of MRI include longer scanning time than CTA and the need for sedation in pediatric patients.

7.6 Vascular Rings

7.6.1 Double Aortic Arch

The double aortic arch (Fig. 7.4) is the most frequent type of vascular ring and sums up about 40 % of the all anatomical varieties.

This vascular malformation consists of the presence of a double aortic arch, one left and one right, both completely patent; generally one of the two arches is larger than the other, and the descending aorta is generally left sided although it may be right sided or positioned in the midline.

In 75 % of the cases, the right arch is dominant, while in the remaining 25 %, the left is the dominant one or there is a balance between the two [7].

This malformation is often associated with other cardiac defects, such as tetralogy of Fallot, truncus arteriosus, or pulmonary atresia.

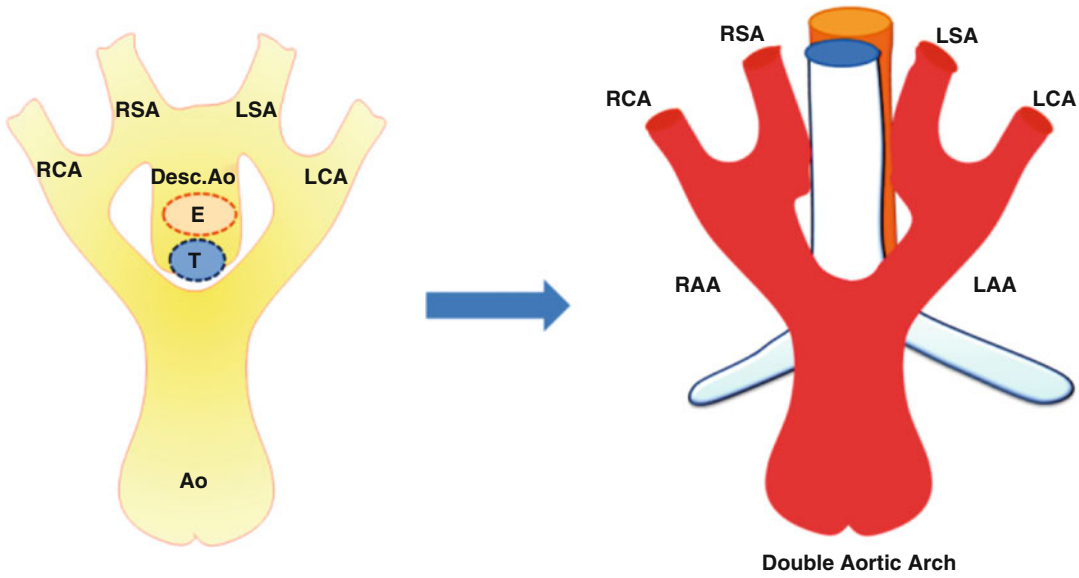

Fig. 7.4 Double aortic arch: persistence of the right and left aortic arch, both completely patent. Legend: *Ao* aorta, *Desc.Ao* descending aorta, *E* esophagus, *T* trachea, *RCA* right carotid artery, *RSA* right subclavian artery, *LCA* left carotid artery, *LSA* left subclavian artery

About 20 % of the cases are associated with chromosomal anomalies such as 22q11.2 deletion syndrome (CATCH 22) [7].

The clinical presentation can include respiratory and/or digestive symptoms, and the surgical correction includes the resection of the nondominant arch. In case of a balanced double aortic arch, it is preferred to maintain the right-sided arch. The surgical approach through a thoracotomy is usually performed on the opposite side of the dominant arch.

Surgical correction usually allows resolution of the symptoms, despite in some cases clinical improvement is not immediate but delayed in time, due to the persistence of the constriction previously established on the esophagus and trachea.

7.6.2 Right Aortic Arch, Aberrant Left Subclavian Artery, and Left Ligament (RAA-ALSA)

The presence of a right aortic arch with left subclavian artery arising from a Kommerell diverticulum is the second most common vascular ring and represents about 30 % of cases, and in 5–10 % of cases, it can be associated with other congenital cardiac abnormalities such as tetralogy of Fallot or ventricular septal defects (VSD).

The embryological development is related to a break occurring between the left common carotid artery and the left subclavian artery of the left-sided fourth brachial branch. The subsequent anatomy is a right aortic arch with aberrant left subclavian artery (ALSA) (Fig. 7.5). The ALSA usually takes origin from the Kommerell diverticulum, representing the remnant of the right dorsal aorta and, in some cases, can create a posterior compression of the esophagus.

This is the spot where the vestige of the ligamentum arteriosum connects the aortic arch to the ipsilateral pulmonary artery forming a complete vascular ring.

The existence, at once, of a bulky Kommerell diverticulum and the residual ligamentum arteriosum causes an important compression above the posterior esophageal wall, showing up with swallowing difficulty.

With the body growing, Kommerell diverticulum can evolve into an aneurysmatic formation, causing dysphagia in adulthood and could be the source of life-threatening collateral events, such as aortic dissection or wall rupture. The risks of rupture and aortic dissection are strictly related to the dimension of the diverticulum [8, 9].

The surgical repair is indicated in the presence of clinical symptoms and consists of the resection

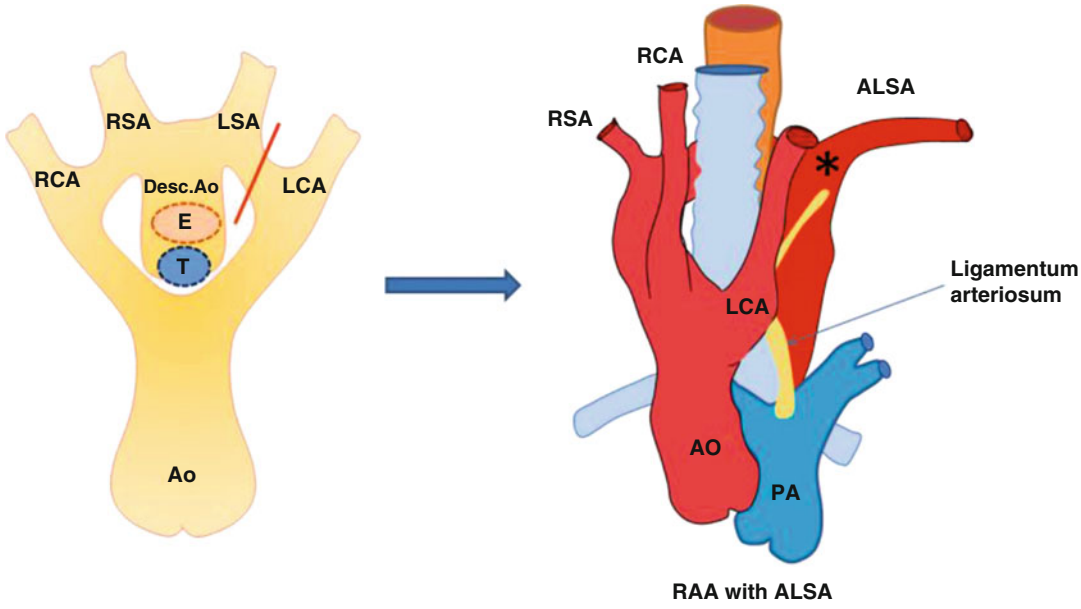

Fig. 7.5 Right aortic arch with aberrant left subclavian artery. Legend: *Ao* aorta, *PA* pulmonary artery, *RCA* right carotid artery, *RSA* right subclavian artery, *LCA* left carotid artery, *ALSA* aberrant left subclavian artery; * = Kommerell diverticulum

of the ligamentum arteriosum, resection of Kommerell diverticulum, and reimplantation of the left aberrant subclavian artery to the left common carotid artery. This surgery can be complex due to the extreme fragility of the diverticulum wall.

In adult patients when complete resection of the aneurysmatic segment of the thoracic aorta is required, the surgery must be performed with the aid of cardiopulmonary bypass with or without hypothermic circulatory arrest. In these cases, the surgery is very challenging and includes a significant risk of death (12 %) and paraplegia (4 %).

7.6.3 Right Aortic Arch with Mirror Image of the Arch Vessels and Retroesophageal Ligamentum (RAA)

This type of vascular ring named right aortic arch with *mirror image anatomy* results from the persistence of the fourth right aortic arch and obliteration of the left one. When reabsorption takes place in the posterior portion of left dorsal arch, behind the origin of the left subclavian artery, the consequent vascular anatomy will be characterized by a right aortic arch with the left brachiocephalic artery (BCA, Fig. 7.6).

Usually this anatomy is asymptomatic, and it does not form a vascular ring. However in rare cases, the presence of ligamentum arteriosum can pull back the BCA realizing an anterior tracheal compression. The RAA could be associated with conotruncal cardiac anomalies such as tetralogy of Fallot or double outlet right ventricle (DORV) and could be associated with genetic syndromes as CATCH 22.

Surgery is indicated only in cases when a tracheal compression is evident. In case of tracheal compression by the right ascending aorta or anomalous innominate artery, it may be performed as a simple aortopexy pulling the aorta toward the sternum, associated with the resection of ligamentum arteriosum.

7.6.4 Left Aortic Arch with Aberrant Right Subclavian Artery (LAA-ARSA)

The presence of a left aberrant subclavian artery is definitely the most common anomaly associated with the left aortic arch. This anomaly occurs

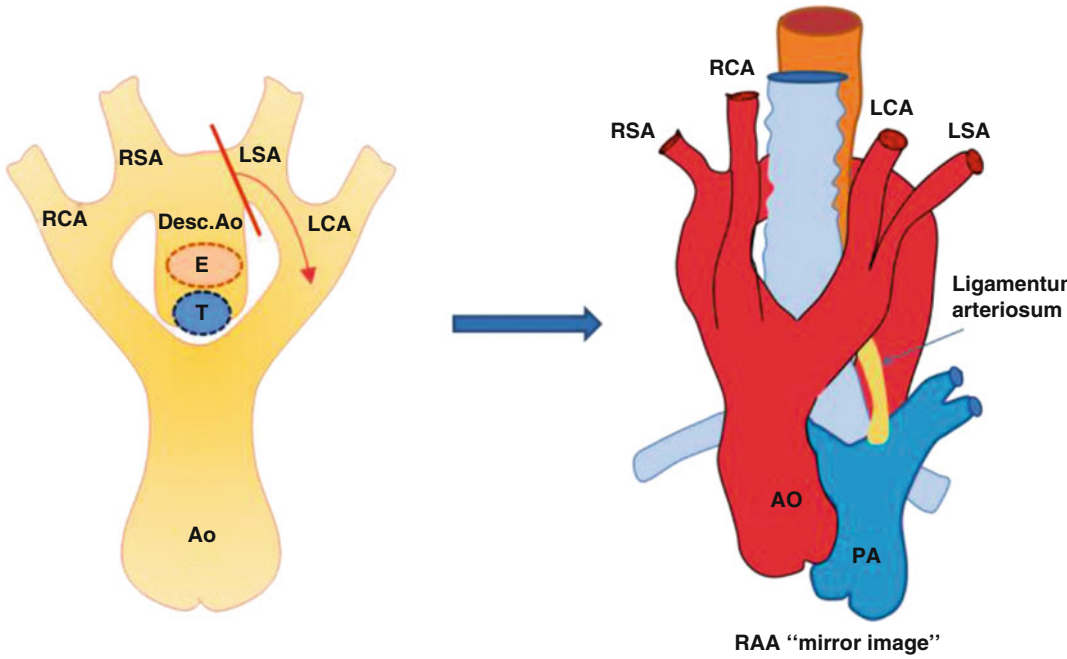

Fig. 7.6 Right Aortic Arch (RAA) "mirror image". Legend: *Ao* aorta, *PA* pulmonary artery, *RCA* right carotid artery, *RSA* right subclavian artery, *LCA* left carotid artery, *LSA* left subclavian artery

when the portion of the right arch, included between the origin of the right subclavian artery and the right common carotid artery, undergoes reabsorption; it usually is not associated with a vascular ring (Fig. 7.7).

Sometimes the right aberrant subclavian artery arises from a Kommerell diverticulum, leading, in this case, to the formation of a vascular ring. This variant is rarely symptomatic, and symptoms usually depend on a voluminous diverticulum causing swelling discomfort or difficulty.

Occasionally, its presentation is associated with other cardiac defects, among which coarctation of aorta, hypoplastic left heart syndrome, tetralogy of Fallot, or a ventricular septal defect. Surgical repair is rarely indicated and consists of the resection of the ligamentum arteriosum and, if needed, resection of the Kommerell diverticulum. In many cases, the surgical treatment was found to be completely unsuccessful because the majority of these patients continued to have symptoms.

7.6.5 Pulmonary Sling

This anomaly is characterized by an unusual drift of the left pulmonary artery, climbing over the right bronchus, passing behind the trachea and in front of the esophagus, heading to the left hilum (Fig. 7.8).

The left pulmonary artery is often hypoplastic and smaller than the right artery, which can eventually be enlarged because of the volume overload; the small size of the left pulmonary artery may help to explain the high incidence of anastomotic problems that have been observed in the past with attempts to reimplant it.

Approximately 50 % of patients with a pulmonary artery sling have complete tracheal rings, that is, the posterior membranous component of the trachea is absent, and the tracheal cartilages, rather than being U shaped, are O shaped.

It usually presents as isolated malformation, but it can be associated with other congenital heart diseases like tetralogy of Fallot.

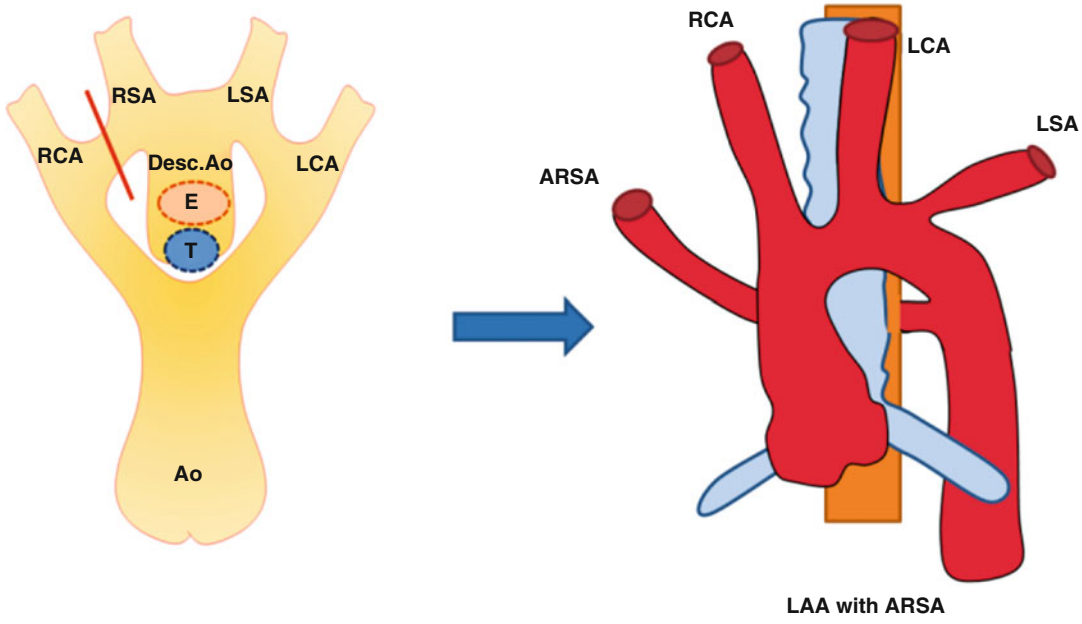

Fig. 7.7 Left aortic arch with aberrant right subclavian artery. Legend: *RCA* right carotid artery, *ARSA* aberrant right subclavian artery, *LCA* left carotid artery, *LSA* left subclavian artery

The clinical scenario is dominated by the compression of the distal part of the trachea associated with severe tracheal and/or bronchial hypoplasia and stenosis. Respiratory symptoms predominate because of the direct tracheal compression and are essentially the same respiratory symptoms as those described for vascular rings. In case of severe airway obstruction, it is necessary to establish a preoperative ventilator support or occasionally an extracorporeal membrane oxygenation (ECMO). Symptoms of esophageal compression are rarely present.

Surgical repair should be performed in those patients showing respiratory obstructive difficulty and consists of a relocation and reimplantation of the left pulmonary artery anterior to the trachea.

Additionally, surgery on the trachea can be required, because of the narrowing provoked by the compression around the portion surrounded by the sling; tracheal surgical repair can complicate the correction, increasing the risk of unsuccessful outcome. For the pulmonary artery relocation and tracheal repair, surgery is per-

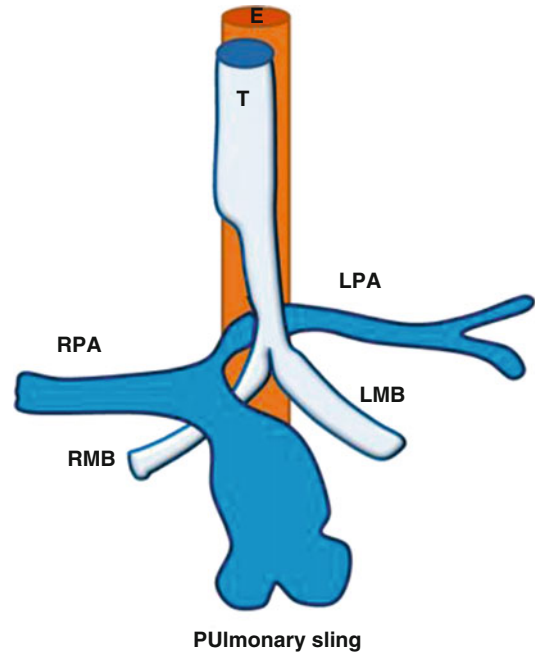

Fig. 7.8 Pulmonary artery sling. Legend: *T* trachea, *E* esophagus, *RPA* right pulmonary artery, *LPA* left pulmonary artery, *RMB* right main bronchus, *LMB* left main bronchus

formed by central sternotomy, and extracorporeal circulation may be required in order to maintain an adequate oxygenation of the patient.

The crucial point of this correction is represented by tracheal surgery. The experience in the treatment of tracheal stenosis remains limited, and the size criteria for tracheoplasty have not been clearly established. Many techniques have been proposed with related advantages and disadvantages.

Direct resection of the stenotic segment and direct anastomosis are ideal for locally limited stenosis. Tracheal reconstruction using pericardial patch is an option for more extended narrowing but currently is less used due to associated problems like patch collapse or excessive phenomenon of granulation in the midterm follow-up. Nowadays the technique of choice is considered the slide tracheoplasty. This technique, firstly introduced by Tsang [10] and popularized by Grillo [11], is used currently both for localized and diffuse stenosis. The main advantage of this technique is the possibility of avoiding the use of graft material with immediate stability of the trachea, reducing the ventilation time and consequently the excessive granulation process.

The trachea is divided transversely at the midpoint of the narrow segment. Subsequently, a longitudinal incision of the proximal and distal segment of the divided trachea is performed. Such incision is carried across opposite (facing) walls of the two segments, in order to complete a sliding oblique anastomosis to restore a final large working diameter.

In some cases with associated tracheobronchomalacia, it may be necessary, after repair, to stabilize the trachea by placing endotracheal stents.

7.7 Treatment and Results

7.7.1 Vascular Rings

Surgery is recommended in all symptomatic patients, especially those with severe respiratory distress. In those patients, the surgery is mandatory in order to avoid tracheobronchial damage or unexpected and graves events, like sudden death.

Asymptomatic patients do not require surgical repair, unless surgical treatment of an associated congenital heart defect is necessary.

The surgical approach depends on the type of vascular rings or sling, and it may be performed by sternotomy, thoracotomy, or in specific cases video-assisted thoracoscopic surgery (VATS).

Nowadays the surgical correction of vascular rings can be carried out with low mortality and morbidity, as described by Backer and coll [12]. In a cohort of 209 patients, who underwent surgery between 1949 and 2003; there was no operative mortality since 1959, and morbidity was mainly related to airway issues: four patients (2%) required late aortopexy for recurrence of airway symptoms.

Recently Ruzmetov and coll. reviewed their experience in the treatment of vascular rings from 1970 to 2008 in 183 patients [13]. There was no intraoperative mortality, but three patients died within 30 days of surgery; mean follow-up was 9 ± 8.3 years, and overall survival was 96% at 35 years [13]. Overall freedom from reoperation was 100% at 35 years, and 75% were free from compressive symptoms within 1 year of the operation; none of the patients showed any evidence of recurrent vascular ring anomalies at the last follow-up [13].

7.7.2 Pulmonary Sling

In cases of diagnosis of pulmonary sling, the repair is mandatory, in order to avoid the progressive damage of the trachea and lung and dangerous asphyctic spells. The results of pulmonary sling surgery without tracheal repair are excellent with low mortality and morbidity, and also the repair of associated anomalies can be performed with no added mortality. Yong and colleagues reported no mortality in nine patients who undergone pulmonary sling surgery alone and 25% of mortality in patients with associated airways surgery [14].

The most important factor to achieve good results in surgery of pulmonary sling seems to be the diameter of the trachea. Huang and colleagues reviewed a small number of patients who underwent treatment of pulmonary sling

showing no mortality in the cohort of patients with internal tracheal diameter larger than 3 mm and in which it was not associated a tracheal surgery [15].

Surgical treatment of tracheal stenosis has been reported having different results due to the complexity of the anatomy (length of the narrowing and diameter of stenotic segment), the presence of tracheomalacia, and the type of surgical techniques.

Backer and colleagues, in their series of 28 patients treated with pericardial patch tracheoplasty, reported 6 % of early mortality and 18 % of late deaths [16].

Similar results (11 % of mortality) are reported by Fanous and colleagues in their series of 26 patients who undergone patch tracheoplasty [17].

Recently, several reports demonstrated that slide tracheoplasty is the technique of choice because it is burdened by a lower mortality and postoperative airway complications. Manning and colleagues reported an operative mortality of 2.5 % after slide tracheoplasty in 80 patients operated between 2001 and 2009 [18].

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