Pediatric Surgery International (2018) 34:701–713
https://doi.org/10.1007/s00383-018-4291-8

REVIEW ARTICLE

Congenital tracheal malformations

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Accepted: 22 May 2018 / Published online: 30 May 2018

Abstract
Congenital malformations of the trachea include a variety of conditions that cause respiratory distress in neonates and infants. A number of anomalies are self-limiting while others are life-threatening and require immediate therapy. The prevalence of congenital airway malformations has been estimated to range between 0.2 and 1 in 10,000 live births. The most frequent congenital tracheal malformations are: tracheomalacia, congenital tracheal stenosis, laryngotracheal cleft and tracheal agenesis. The management of congenital tracheal malformations is complex and requires an individualized approach delivered by a multidiscipilinary team within centralized units with the necessary expertise.

Keywords Congenital malformations · Tracheomalacia · Tracheal stenosis · Aortopexy · Tracheoplasty · Tracheal stents

Introduction

Congenital malformations of trachea are uncommon but a challenging field in pediatric surgery due to the complexity of the disease. Tracheomalacia and tracheal stenosis are the most common congenital abnormalities of the pediatric airway. Less common tracheal anomalies include laryngotracheaoesophageal clefts and tracheal agenesis. Pediatric airway surgery is challenging and required careful diagnostic investigation, and urgent and demanding reconstructive treatments.

Tracheal agenesis

Tracheal agenesis is very rare, occurring in 1 in 50,000 to 1 in 100,000 live births and usually results in fatal outcome [1]. The cervical trachea is usually absent. The bronchus or the carina is connected to the esophagus. Floyd proposed an anatomic classification in three types [2]: type I; representing the 20%. There is agenesis of the upper trachea. Bronchus is normal. There is a tracheoesophageal fistula. Type II; the most frequent representing 60% of all the cases reported. There is a complete tracheal agenesis. Bronchus are normal and there is a fistula between the carina and esophagus. Type III; the bronchus arises from the esophagus separately (Figs. 1, 2).

The presence of a tracheoesophageal or bronchoesophageal fistula is important for salvage treatment of a critically ill neonate with tracheal agenesis, because it allows esophageal intubation and mechanical ventilation. Esopahgeal intubation is lifesaving. Tazuke et al. [3] reported four patients with tracheal agenesis with long-term survival after airway and esophageal reconstruction. Densomore et al. [4] have modified this surgical approach to save a particularly high-risk patient with Floyd II tracheal agenesis by neonatal esophageal trachealization and esophagocarinoplasty.

Laryngotracheoesophageal cleft

The laryngeal and tracheal cleft is an abnormal communication between the trachea and esophagus. The extension of communication is variable, from a cleft at the level of the larynx exclusively to a wide communication between trachea and esophagus that may extend even to the bronchus. When cleft extends below the cricoid cartilage is

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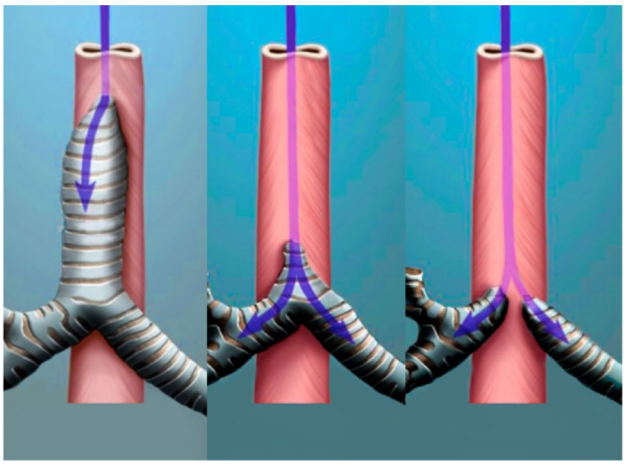


Fig. 1 Tracheal agenesis type I, II, III

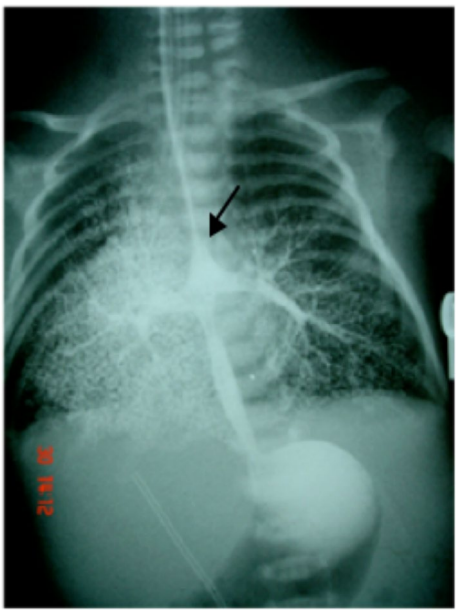


Fig. 2 Tracheal agenesis. Upper gastrointestinal contrast study. Both bronchus are connected to the esophagus through a fistula (arrow)

called laryngotracheo esophageal cleft. According to Benjamin and Inglis, clefts are classified according to the severity into four types (I–IV) [5–7] (Fig. 3).

The diagnosis is based on the suspicion index. Sometimes, the symptoms are nonspecific, but most have associated respiratory symptoms. Characteristically cough or early choking during feeding is secondary to aspiration. Patients may present with airway and/or impairments [8]. Laryngomalacia and tracheomalacia are frequently associated.

The definitive diagnosis must be performed using an endoscopic evaluation with rigid scope under general anesthesia. Type I laryngeal cleft generally does not need to be repaired. The majority of patients with type I are asymptomatic. If there is no clinical and radiological pulmonary aspiration, the surgical intervention is not required. Type II–III and IV require surgical repair (Fig. 3). Type IV repair is extremely complex and associated with severe tracheomalacia requiring in many cases ventilation support, tracheostomy and even gastrostomy.

Technical aspects

Endoscopic repair is indicated for type I symptomatic and II even for some types of III (Fig. 4 a–c). The approach of choice for III–IV is open trans-tracheal through the neck and even combine approach with a mid-sternotomy (Figs. 5, 6, 7). All types of clefts can be repaired by this approach. A two-layer repair is essential, dissecting the tracheal and esophageal mucosal layer and performing a closing of both mucosal separately. It is recommended to interpose synthetic fibrin, perichondrium or periosteum between the two layers [9–15] (Figs. 5, 6, 7). In the case of larger cleft (IV), support with ECMO during repair is recommended [16].

Congenital tracheal stenosis

Congenital tracheal stenosis (CTS) is a rare condition with an estimated incidence of 1 in 64,500 births [17]. CTS is a true embryological abnormality of the tracheal skeleton, with the presence of complete tracheal rings along the stenotic segment and determine a fixed narrow tracheal lumen. In the normal trachea, the cartilaginous rings are incomplete or horseshoe shaped [18]. In most of the cases, there is not a transition zone between the normal and complete rings, only in few cases it is possible to identify a transition tracheal segment with rings from normal horseshoe shape to a complete (Figs. 8, 9, 10, 11a, b). CTS is associated with a cardiovascular abnormality in 69% of the cases. The most common vascular associated is the left pulmonary artery sling found in 44% (Figs. 12, 13). Tracheal origin of the right upper lobe bronchus is found in 50% of the cases [19].

Symptoms

In general, symptoms start in a few days after birth and are associated with a severity of the stenosis more than stenotic length. The gold standard for the diagnosis is the rigid airway endoscopy. Computed tomography (CT) or magnetic resonance images (MRI) are essential to study the associated vascular malformations. The symptoms in CTS are variable and are directly related to the degree of narrowing of the lumen and stenotic length.

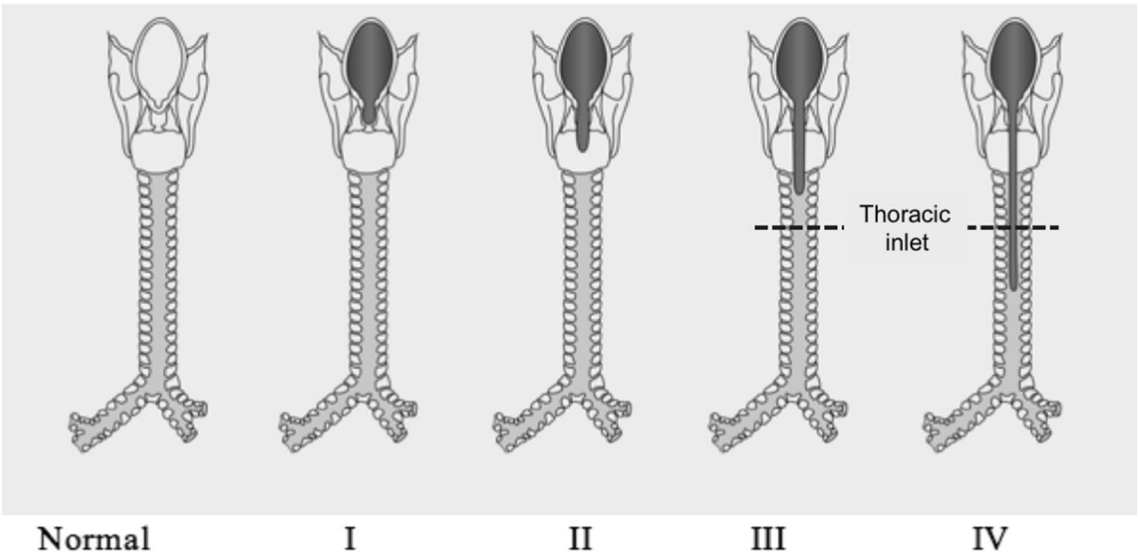


Fig. 3 Laryngeal and tracheal clefts. Benjamin and Inglis classification

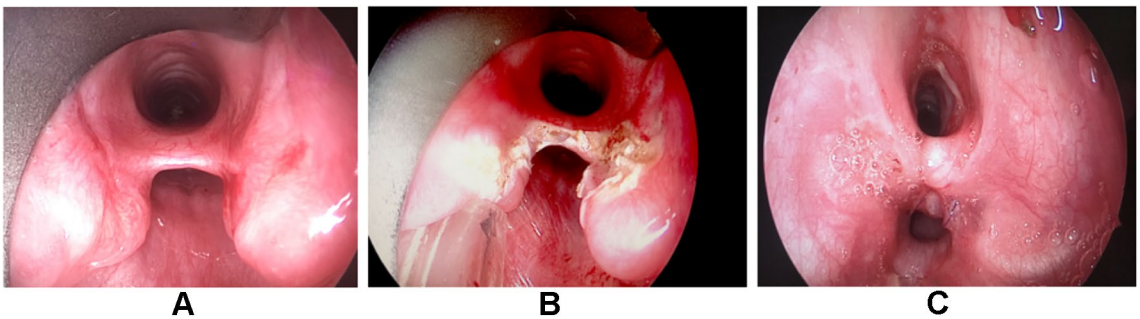


Fig. 4 **a** Preoperative endoscopic view of a type III cleft. Airway and esophagus are connected in the upper level. The distal end of the cleft lies lower than the inferior border of the cricoid plate. **b** Endoscopic cleft repair. The mucosa is refreshed before stitched. **c** After endoscopic repair

Diagnosis

Chest X-ray can suspect the diagnosis of tracheal stenosis, and a bronchography with iso-osmolar contrast can be useful in confirming the diagnosis (Figs. 14a, b, 15). CT with 3D reconstructions and MRI with digital substraction are the radiological studies of first choice for achieving a clear assessment of the relationship between the airway and mediastinal cardiovascular anomalies if present (Figs. 16, 17, 18). The airway endoscopy evaluation is the gold standard for the diagnosis. The presence of complete tracheal rings is characteristic.

Morphological classification

There is a morphological classification for CTS proposed by Grillo long time ago [20] (Fig. 19).

Type I: Long segment tracheal stenosis. It is the most frequent type and usually compromised more than 85% of tracheal length. Only the first to fourth upper rings are normal (Figs. 8, 9, 11).
Type II: Funnel-shaped stenosis.
Type III: Short segment. Stenosis is less than 50% of the tracheal length and is associated with abnormal upper right bronchus (Figs. 14, 17).
Type IV: Characterized by the presence of an abnormal upper right bronchus and a long bridge bronchus (Figs. 16, 18). In this variant of type III, the anomalous right upper lobe bronchus is in the proximity of the carina, and via horizontally branching bronchia a stenotic bridge bronchus connects the proximal trachea to the rest of the lungs [19].

Recently, a new classification was proposed by the Great Ormond Street Hospital based on the bronchial arborization

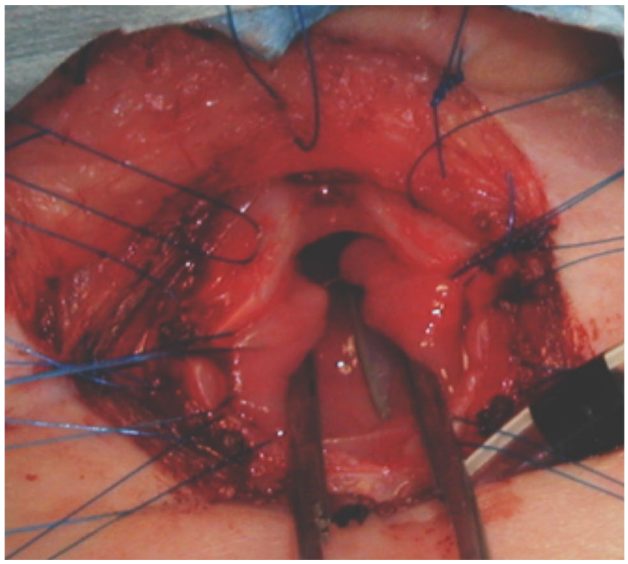


Fig. 5 Type III cleft. Cervical approach. Communication between esophagus and trachea is observed. Esophagus shows nasogastric tube

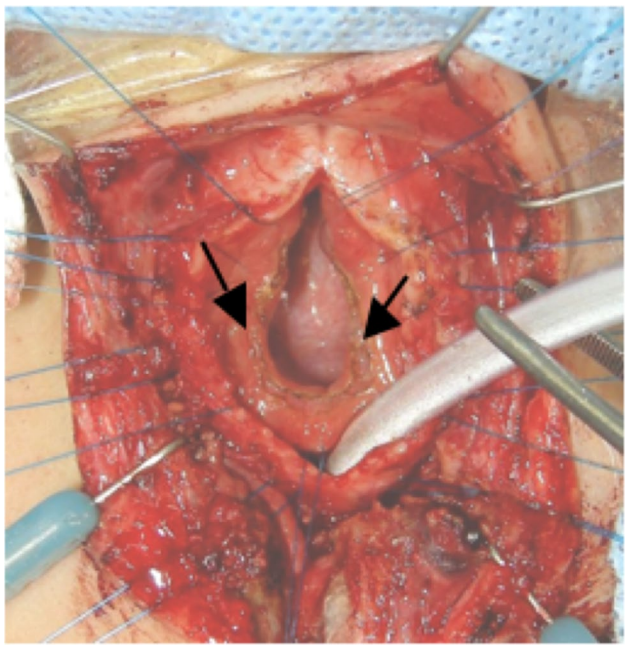


Fig. 6 Larynx and trachea are exposed. The common wall between esophagus and trachea is divided (arrows)

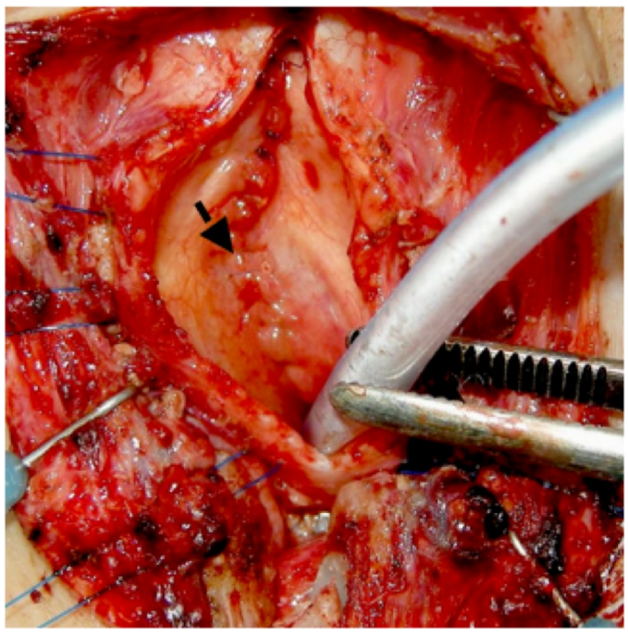


Fig. 7 Esophagus and posterior tracheal wall are closed in two layers (arrow)

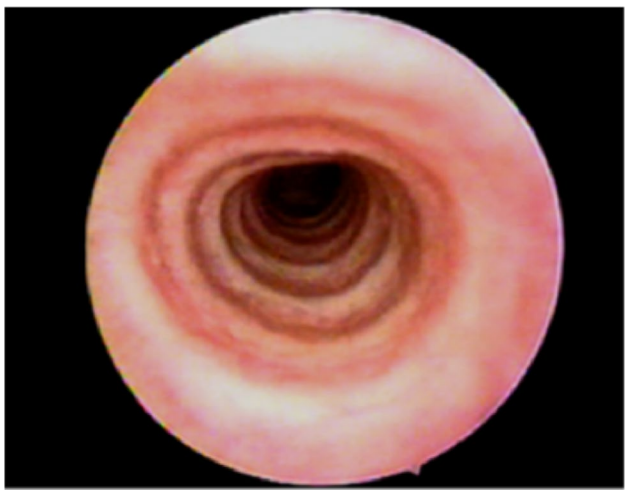


Fig. 8 Congenital complete tracheal rings. Rigid scope view. Complete tracheal rings (360°) can be seen and there is an absence of membranous posterior wall

and complete ring localization and found to be useful for the morphological characterization of CTS [21].

CTS surgical repair

The surgical indication depends on the presence of respiratory symptoms. There is controversy regarding the surgical indication in patients with congenital tracheal stenosis who have mild symptoms. Some studies have shown that the growth of the tracheal diameter over the years might reduce symptoms. It has been suggested that the presence of a pulmonary artery sling is an indication for a surgery even in patients with mild symptoms. Tracheal stenosis and vascular malformation should be repaired at the same time. The artery sling required reinsertion of the left pulmonary artery in the trunk of the pulmonary artery [22, 23].

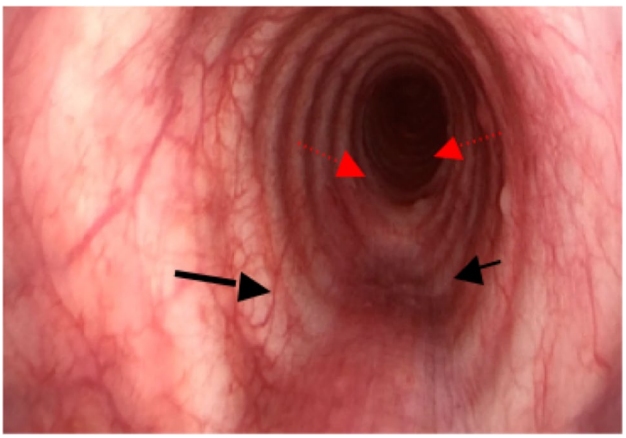


Fig. 9 Long-type congenital tracheal stenosis: rigid endoscopy view. The first four upper rings are normal (arrow) and easily distinguished from circular rings (red interrupted arrow)

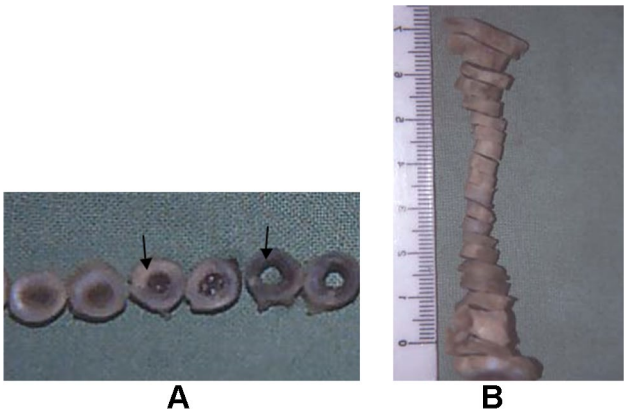


Fig. 11 **a, b** One-year-old patient. Died after a cardiac surgery. In the post-operative airway pathology examination, a tracheal malformation was found. The tracheal rings (black arrows) with a severe stenosis and complete obstruction of the lumen secondary to bleeding

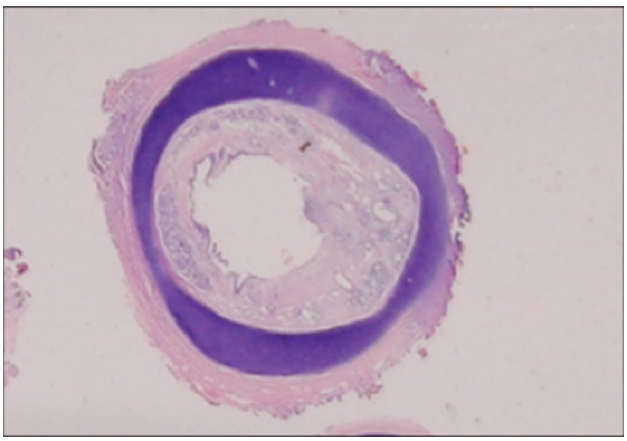


Fig. 10 Histology of a complete tracheal ring. Absence of membranacea posterior wall

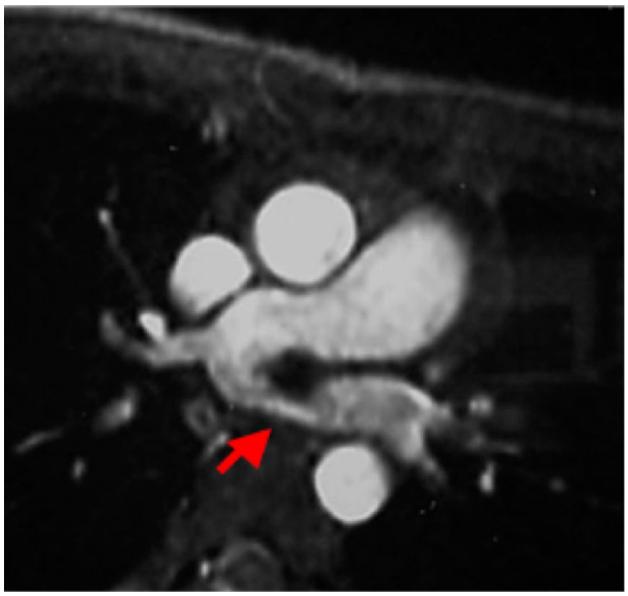


Fig. 12 CT scan with contrast shows a left pulmonary artey sling (LPAS) (arrow). The LPAS is arising from the right and it goes behind the trachea to the left side

Long CTS: surgical techniques

In long CTS, multiple surgical techniques for the correction of this complex abnormality have been developed such as pericardial patch, slide tracheoplasty technique, tracheal autograft technique and cadaveric tracheal homograft. In the last two decades, all the above techniques have been replaced by the slide tracheoplasty technique.

Long CTS: pericardial patch technique

In the past, pericardial patch (Figs. 20, 21) was considered for long stenosis. Actually, this technique is not recommended and has been replaced by the "slide tracheoplasty" technique [20, 22–29].

Long CTS: slide tracheoplasty technique

Slide tracheoplasty technique (Figs. 22a–d, 23) consists of a tracheal transection at the midpoint of stenosis and expansion of both tracheal segments, follow to a slide. Both

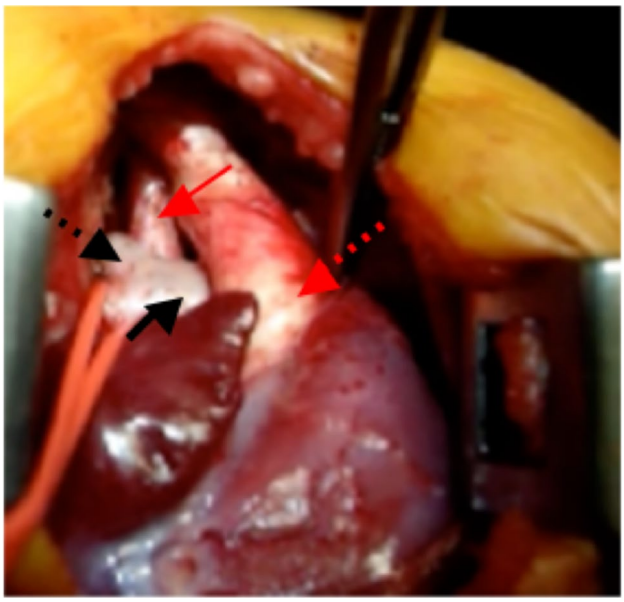


Fig. 13 Intra-operative: right pulmonary artery (black arrow), origin of left pulmonary artery sling (interrupted black arrow), trachea (red arrow), aorta (interrupted red arrow)

tracheal segments follow to a slide. Both tracheal segments are stitched using absorbable or non-absorbable monofilament suture (running or interrupted). The tracheoplasty can be carried out by cervical or thoracic approach. We suggest sternotomy. The long CTS associated with a cardiac or vascular malformation require generally extracorporeal circulation support (Figs. 24, 25) ensuring adequate ventilation and oxygenation while the tracheal lumen is open and the tracheoplasty is performed.

Slide tracheoplasty has become increasingly successful over the past two decades such that long-term survival now exceeds 88%, with normalization of quality of life for patients with non-syndromic-associated CTS [18]. Although slide tracheoplasty can be successfully performed in patients with normal pulmonary anatomy, this procedure in patients with abnormal pulmonary anatomy can be more difficult and may require prolonged mechanical ventilation and the use of ECMO [30].

Short CTS

For short stenosis (less than 1/3 of the tracheal length), a resection and end-to-end anastomosis is the most widespread considered technique (Figs. 26, 27).

Tracheomalacia

Tracheomalacia is an abnormal softness of the tracheal wall due to structural anomalies of tracheal cartilaginous and/or posterior membrane. If the malacia is diffused to the bronchi, it is called tracheobronchomalacia or bronchomalacia if it is limited to bronchus. The condition can be congenital (primary) or acquired (secondary). Others call the tracheomalacia secondary when it is associated with other anomalies. Airway malacia including laryngomalacia, tracheomalacia and bronchomalacia is the most common congenital abnormalities of the pediatric airway and are characterized by increased airway compliance, resulting in excessive dynamic collapse during the respiratory cycle [31, 32].

The incidence is underestimated, as the diagnosis is not easy and some severe cases of congenital tracheomalacia

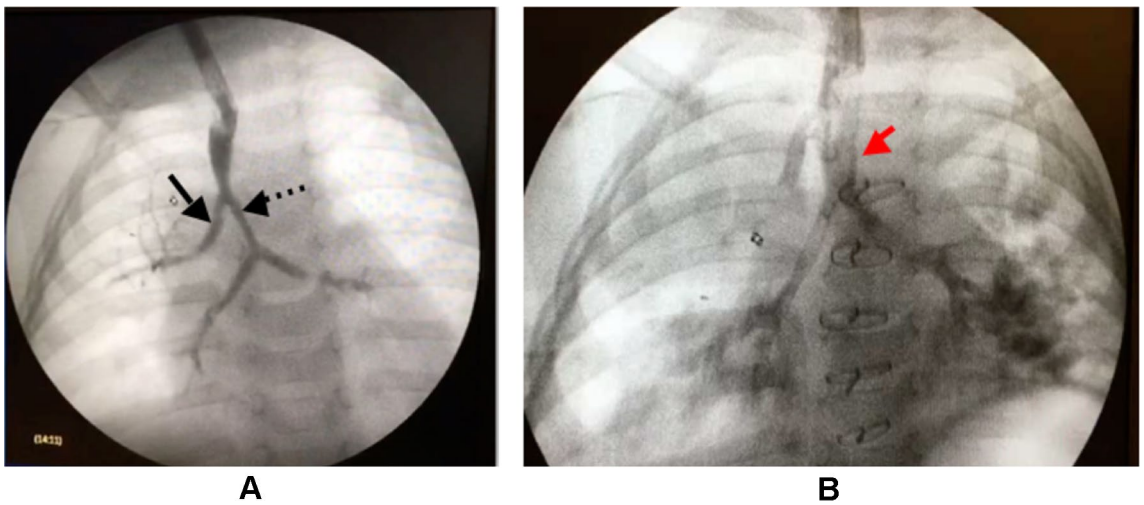


Fig. 14 **a** Bronchography. Preoperative bronchography. A classical type III CTS with the pig or anomalous upper right bronchus (black arrow) arising at the beginning of the stenosis (interrupted arrow). **b** Post-operative bronchoscopy: after a slide tracheoplasty reconstruction: the tracheal lumen is enlarged (arrow). Compare with the previous preoperative picture

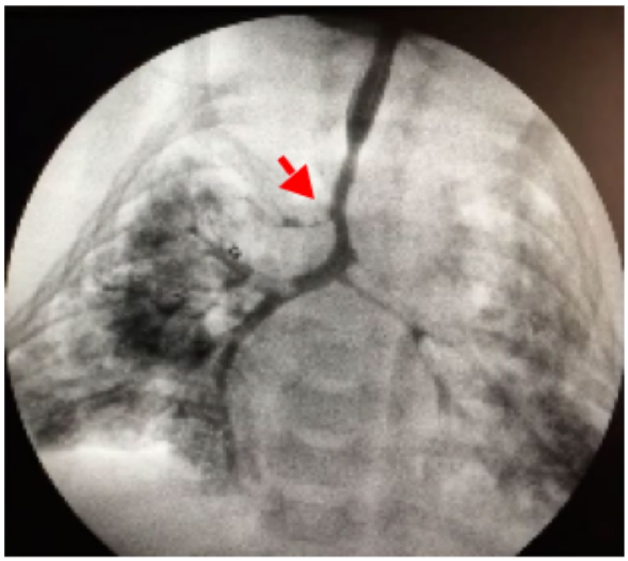


Fig. 15 Bronchography. One-month-old was born with stridor and congenital cardiovascular defect. Type III CTS with the pig bronchus arising in the middle of the stenotic tracheal segment (red arrow)

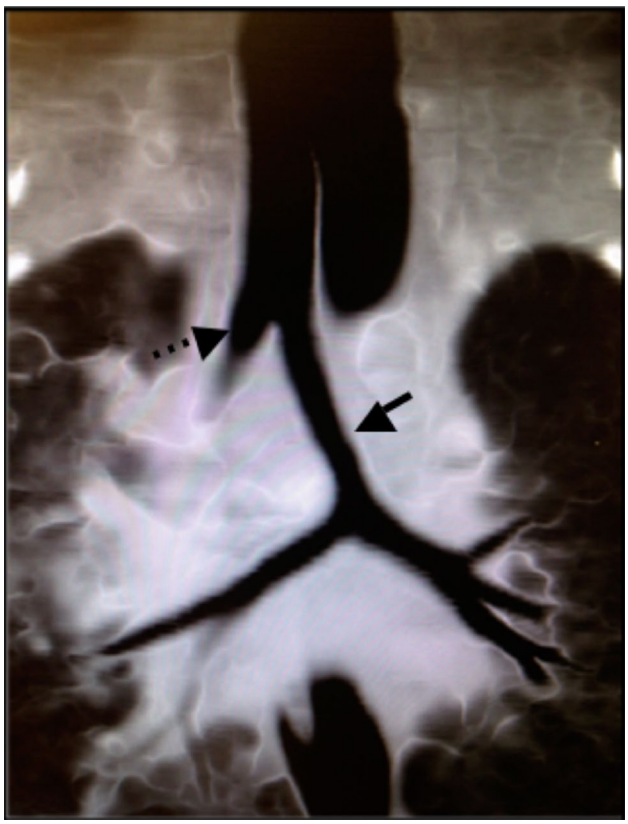


Fig. 16 CT scan shows a congenital tracheal stenosis type IV "bridge bronchus". Segmented arrow: pig bronchus origin. Arrow: bridge bronchus

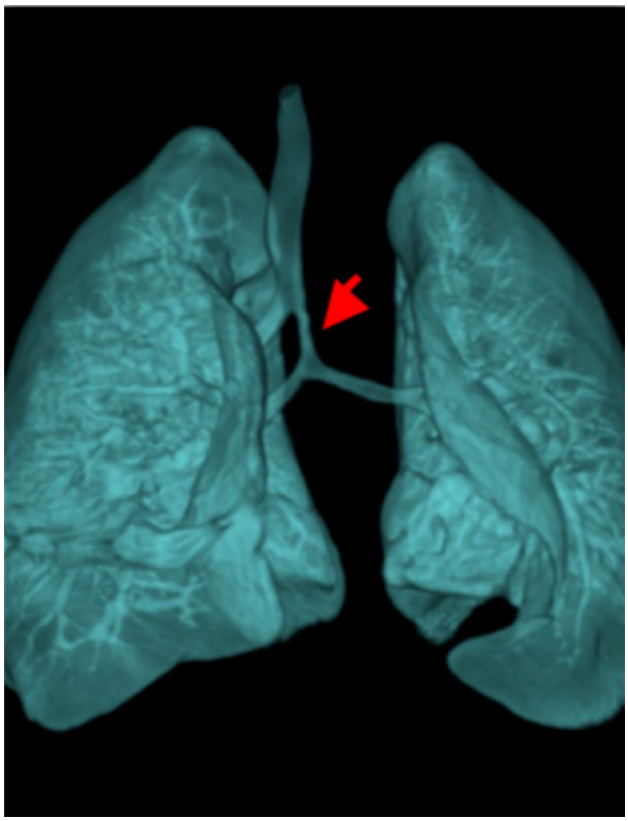


Fig. 17 CT scan. A type III or short CTS. Red arrow shows the stenotic tracheal segment below the pig bronchus origin

may die during early infancy. The embryological development of the trachea from the foregut, a common tubular structure from which esophagus takes origin, provides insights into the pathogenesis of congenital tracheomalacia. The common embryologic origin of the trachea and esophagus explain why tracheomalacia is so often associated with esophageal atresia with trachea–esophageal fistula [1, 33]. At the end of the embryogenesis, trachea and esophagus are fully separated and the wall of the normal trachea is composed of C-shaped cartilage rings anteriorly and of a muscular part (pars membranacea) posteriorly. The pars membranacea could be considered as the residual of the floppy component of the primitive foregut, as well as the esophagus, that is just posterior to the pars membranacea. The ratio among cartilage and floppy part of the normal trachea is around 4.5/1. An abnormal ratio can explain not only tracheomalacia but also other tracheal anomalies: if the pars membranacea is more developed, tracheomalacia usually occurs. If the cartilage is completely absent, a segmental absence of tracheal rings or tracheal agenesis is the result [34, 35].

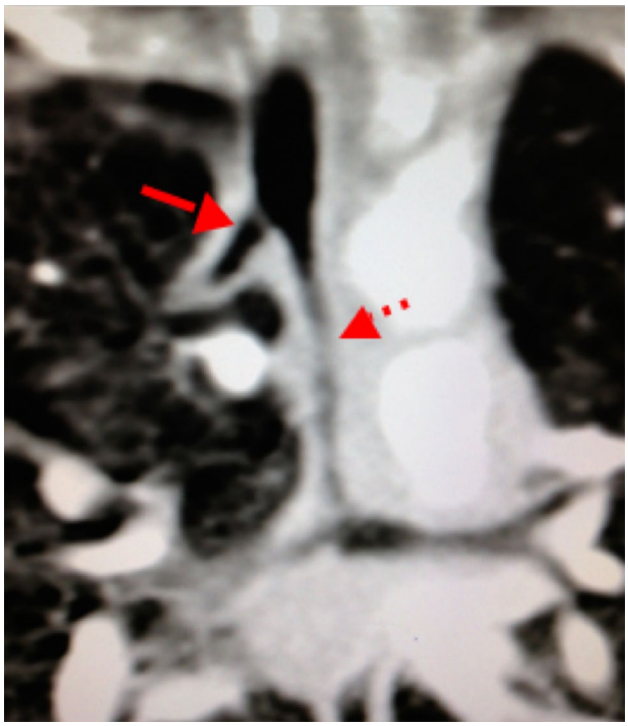


Fig. 18 Type IV CTS, "Bridge type", 4-year-old boy, stridor since birth. A CT scan shows an upper right anomalous bronchus (red arrow) arising upper in the trachea and a long bridge bronchus (red interrupted arrow) connecting the trachea to the lungs

Physiology and pathology

Respiration is a dynamic cycle, with the inspiratory and the expiratory phases. For the pressure differential, during expiration, in particular forced expiration (e.g. cough), the pars membranacea of the intrathoracic trachea intrudes into the tracheal lumen. The cartilaginous framework ensures that this collapse is limited to the physiological 30–35% of lumen reduction. If the dynamic collapse is bigger than 50%, some authors call the condition "Hyperdynamic airway collapse" and differentiate it from tracheomalacia. While hyperdynamic airway collapse is referred to a pars membranacea institution, tracheomalacia is a true collapse or deformation of the cartilage rings. A similar collapse of a floppy airway can be observed during inspiration, but for the opposite pressure balance in this case the collapse will be observed at the level of the cervical trachea [36].

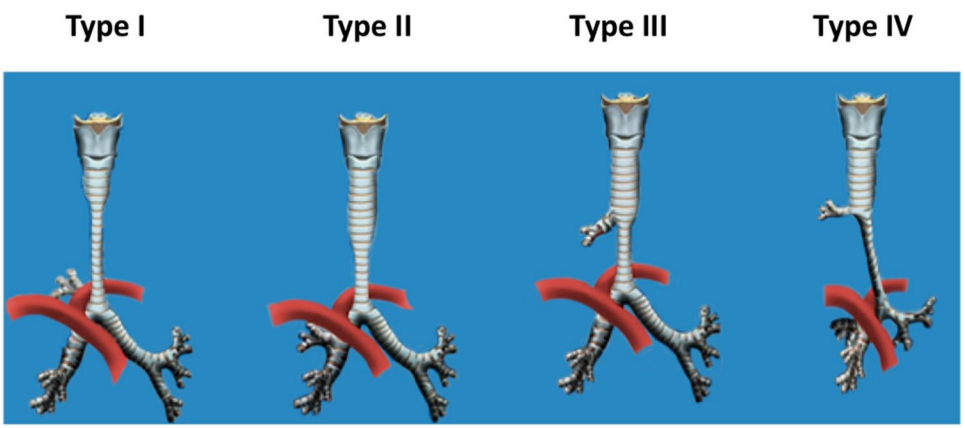
Symptoms

Tracheomalacia causes a large spectrum of respiratory symptoms, due to the airway collapse: respiratory sounds, distress with cyanosis, apnea or acute life-threatening events (ALTE), failure to extubation, exertional dyspnea; or to reduction of secretion clearance: repeated and prolonged infections, lung atelectasis, bronchiectasis. The severity of the symptoms can be very different, depending on the collapse severity and extension. Diffuse and severe tracheomalacia is life-threatening, while mild cases are often self-limiting and spontaneously improving.

Diagnostic evaluations

Diagnosis of tracheomalacia is classically based on dynamic trachea-bronchoscopy, with direct observation of the tracheal collapse during spontaneous breathing. Endoscopy remains the gold standard diagnostic tool; however, it requires general anesthesia, so efforts have been made to find less-invasive alternatives. The aim of both examinations is to evaluate the percentage of reduction of airway lumen and the sites involved (upper, middle, distal trachea, proximal and peripheral bronchi). In many patients, both investigations are performed, as both have high sensibility and specificity and complimentary for evaluating different aspects: dynamic CT is essential or superior to study associated vessel anomalies, trachea diverticula, peripheral bronchi and lung parenchyma, while endoscopy allows to evaluate vocal cord function, proximal airway, other associated anomalies, as clefts or trachea-esophageal fistulas [37].

Fig. 19 CTS classification



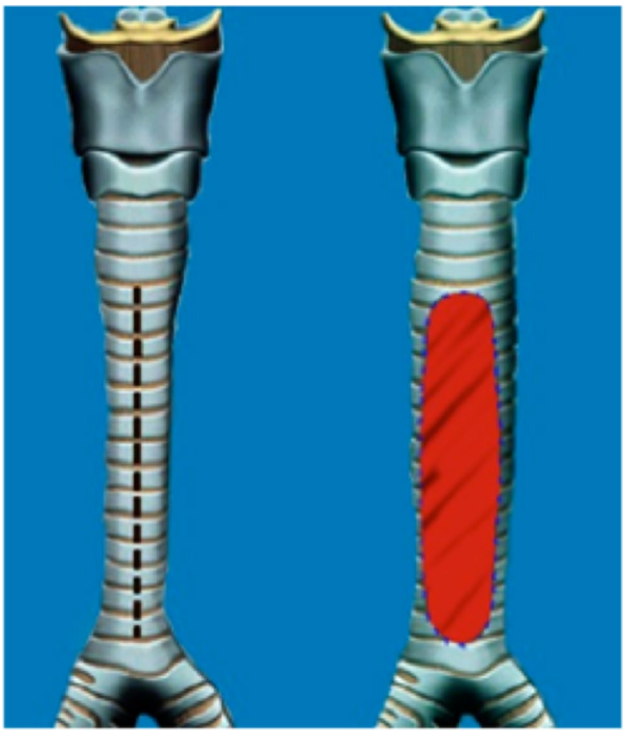


Fig. 20 Pericardial patch enlargement technique

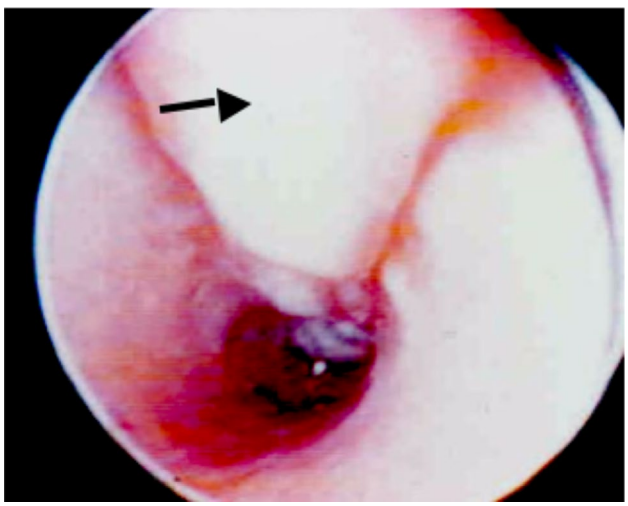


Fig. 21 Pericardial patch enlargement. Post-operative endoscopic view. The patch (arrow) is stitched in the anterior wall after the anterior tracheal wall has been longitudinally opened

Treatment

Tracheomalacia can improve spontaneously, but in some patients the treatment is recommended for the severity of the symptoms or to reduce long-term lung sequelae. As the respiratory status and associated comorbidities differ so much in tracheomalacia patients, the treatment should be individualized choosing one or more among the multiple options of treatment existing. Unfortunately, a Cochrane review failed to demonstrate the efficacy of a single treatment on the others for tracheomalacia [38]. There is no consensus on the optimal treatment of severe tracheobronchomalacia, which can be associated with a mortality rate as high as 80% [39]. We can distinguish two groups of treatments: conservative and surgical. The conservatives are represented by pharmacological tools aiming to treat infections, gastroesophageal reflux, or improve the tonicity of the pars membranacea. An important conservative treatment is continuous positive pressure ventilation (CPAP) that maintains the airway open. In between conservative and surgical treatments, semi-invasive measures can be considered. Tracheotomy is usually a relatively simple procedure, that allows to manage tracheomalacia. The cannula inserted through the tracheostoma allows maintaining the floppy trachea open. Another option is to insert a stent. Many different airway stents are available in the market. They can be inserted in the trachea or in the bronchus. The types of available stents are mainly metallic (Fig. 28) (palmaz, nitinol and wallstents). Nitinol and wallstents are self-expanding. They can be inserted in the tracheal or bronchial lumen. Once on-site they can stay for a long time or eventually be removed. In our opinion, a permanent non-absorbable stent should be avoided in the child, as it will become inadequate in the growing trachea or it will cause granulation that will require repeated endoscopic procedures. The ideal stent for the pediatric airway is absorbable or removable. A study comparing stents and surgery for tracheomalacia found that the results were similar but stents were associated with more morbidity and mortality [40–44].

Absorbable stents

Absorbable stents made of PDS (Fig. 29) are becoming in our experience the best choice especially when tracheomalacia is expected to improve with age. Though sometimes inducing granulations, their net shape allows ciliary movement and clearance of secretions, and in 3–4 months, they are completely dissolved. Among surgical treatment, for short and severe tracheomalacia (e.g. peristomal malacia) a resection of the floppy segment of trachea and anastomosis usually works very well. For longer forms of tracheomalacia, a possible option is to reinforce the airway with an exoskeleton. Absorbable miniplates, rapisorb (Depuy Synthes, Johnson & Johnson) are available in the market and can be attached to the tracheal wall, making it stiffer and avoiding its collapse. This approach is technically simple as the airway is not opened. The plates may be given the appropriate shape by soaking them in hot water [45].

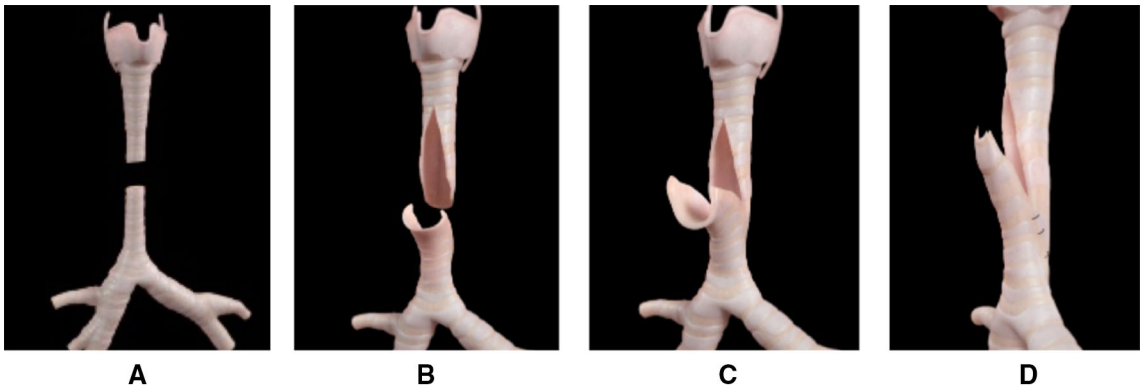


Fig. 22 **a** Slide tracheoplasty technique. Trachea is divided in the stenotic midpoint. **b** Slide tracheoplasty technique. After mid point division, both segments are enlarged. The upper segment is divided anteriorly and the inferior segment posteriorly (as described originally by goldstraw, London). **c** Slide tracheoplasty technique. One segment is slided into the other and stiched. **d** Slide tracheoplasty technique. After segments are slided anastomosis is performed with running PDS or prolin suture

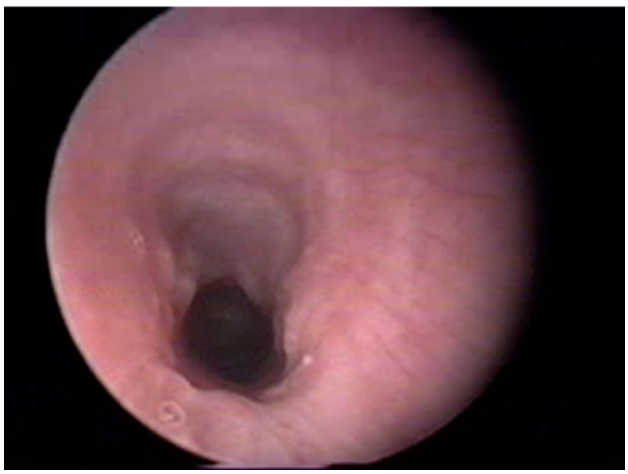


Fig. 23 Post-operative endoscopic view after slide tracheoplasty. The classical "eigth shape" anastomosis

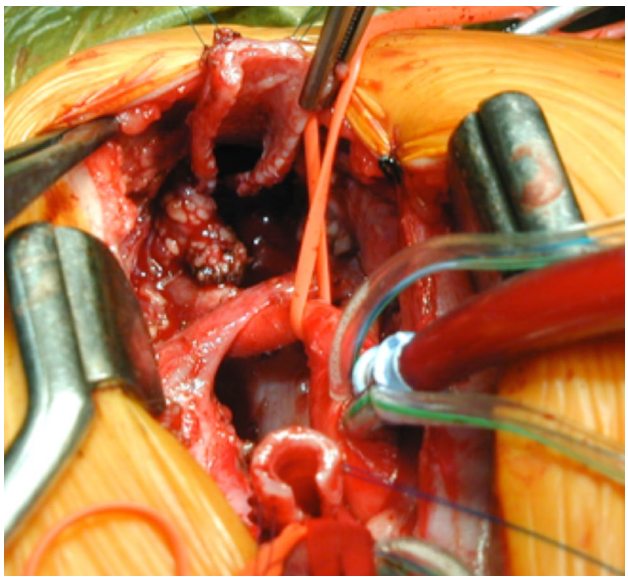


Fig. 24 Tracheoplasty under cardiopulmonary bypass in patient with long segment stenoses. Both segments are already divided

Aortopexy

Aortopexy is one of the most popular surgical treatment of tracheomalacia. The principle of aortopexy is to move anteriorly the aorta and attach it to the sternum through transternal stitches. The trachea will be moved together, as it is attached to the aorta with pretracheal fascia that is not dissected from it. Intra-operative tracheoscopy is useful to check the efficacy of the aortic pull-up. The surgeons will knot the stitches on the anterior sternal plate when they will be sure that the stitches are effective to enlarge the tracheal lumen. Aortopexy can be performed through different approaches: mini-sternotomy, left anterior thoracotomy, and right or left thoracoscopy. Torre et al. [46] reported that aortopexy is effective to improve tracheomalacia symptoms in the majority of cases. A novel technique, described by Boston group, has been recently proposed for those cases in which the airway occlusion is mainly due to pars membranacea posterior intrusion: posterior tracheopexy. The principle of this approach is to enlarge the airway lumen (tracheal and/or bronchial) by fixing the pars membranacea to the anterior spinal ligament. Every stitch is guided by an intra-operative bronchoscopy control. To create the space between the spine and the posterior wall of the trachea, esophagus and thoracic duct have to be separated by the airway and pushed to the right side, while the aorta in some cases must be dislocated to the left. Posterior tracheopexy can be performed through a right thoracotomy or thoracoscopic approach (in case of left aortic arch). The procedure can be performed at the same

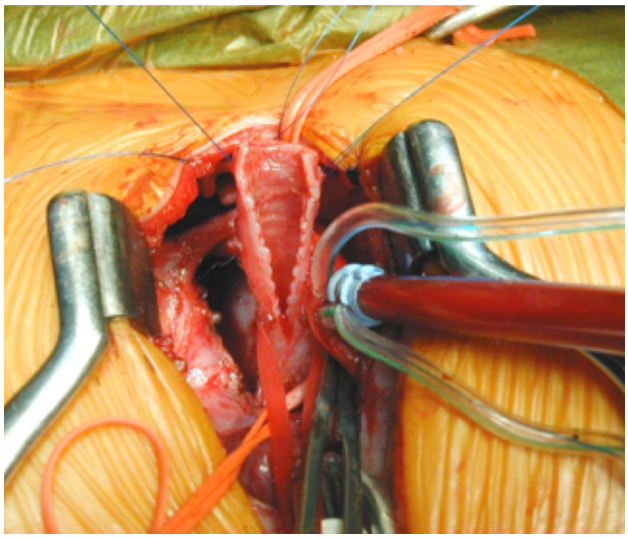


Fig. 25 Slide tracheoplasy. See all the complete rings divided anteriorly in the lower segment (as described by Grillo, US)

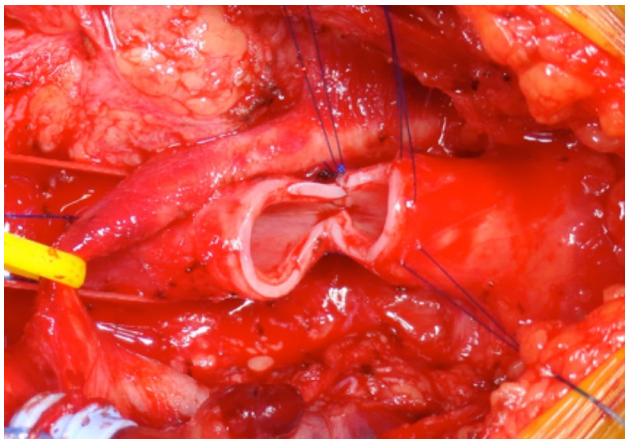


Fig. 26 Tracheal resection and end-to-end anastomosis in a short-type CTS

time as anterior aortopexy if the collapse is both anterior and posterior [47].

Conclusion and future direction

Congenital tracheal malformations include a wide array of anomalies with a broad spectrum of symptoms. A critical and intensive care with a well-disciplined collaboration between pediatric surgeons, neonatologists, radiologists, and anesthetists is required.

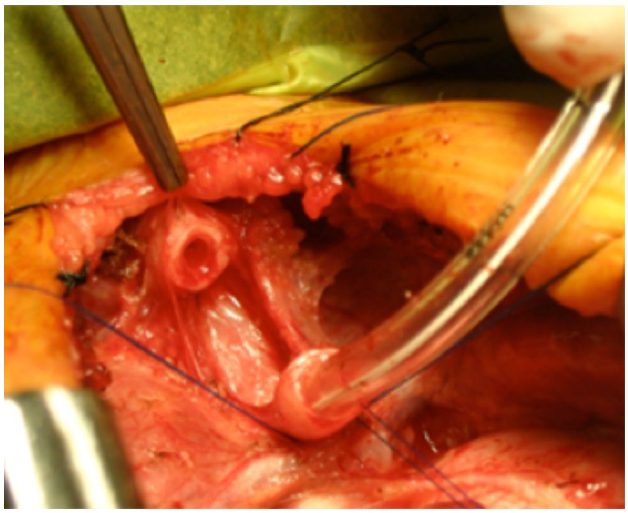


Fig. 27 Short type CTS. Resection and primary anastomosis. Patient is oxygenated with a tracheal tube in the distal segment. No cardiopulmonary support or ECMO is needed

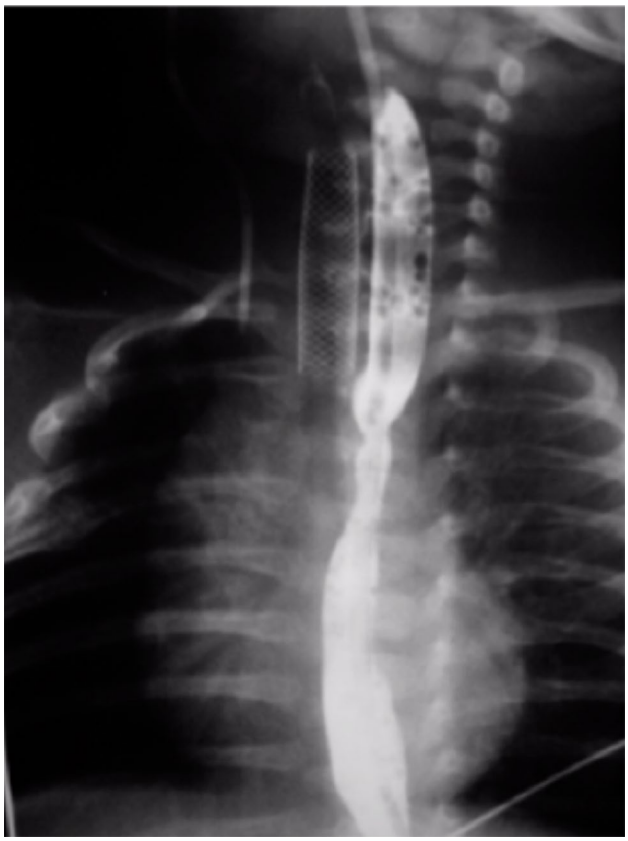


Fig. 28 Chest X-ray, shows a nitinol self-expanding stent in trachea. Patient with severe tracheomalacia associated with esophageal atresia

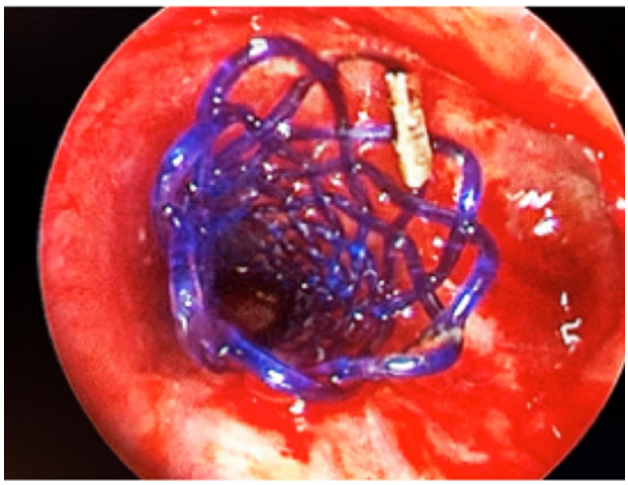


Fig. 29 Absorbable PDS stent in left main bronchus

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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