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ORIGINAL ARTICLE

Surgical treatment for severe pediatric tracheobronchomalacia: the 20-year experience of a single center

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Abstract
Objective: In children with tracheobronchomalacia, surgical management should be reserved for the most severe cases and be specific to the type and location of tracheobronchomalacia. The goal of this study is to describe the presentation and outcomes of children with severe tracheobronchomalacia undergoing surgery.
Methods: Retrospective case series of 20 children operated for severe tracheobronchomalacia at a tertiary hospital from 2003 to 2023. Data were collected on symptoms age at diagnosis, associated comorbidities, previous surgery, age at surgery, operative approach, time of follow-up, and outcome. Surgical success was defined as symptom improvement.
Results: The most frequent symptoms of severe tracheobronchomalacia were stridor (50 %), cyanosis (50 %), and recurrent respiratory infections (45 %). All patients had one or more underlying conditions, most commonly esophageal atresia (40 %) and prematurity (35 %). Bronchoscopy were performed in all patients. Based on etiology, patients underwent the following procedures: anterior aortopexy (n = 15/75 %), posterior tracheopexy (n = 4/20 %), and/or posterior descending aortopexy (n = 4/20 %). Three patients underwent anterior aortopexy and posterior tracheopexy procedures. After a median follow-up of 12 months, 16 patients (80 %) had improvement in respiratory symptoms. Decannulation was achieved in three (37.5 %) out of eight patients with previous tracheotomy. The presence of dying spells at diagnosis was associated with surgical failure.
Conclusions: Isolated or combined surgical procedures improved respiratory symptoms in 80 % of children with severe tracheobronchomalacia. The choice of procedure should be individualized

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and guided by etiology: anterior aortopexy for anterior compression, posterior tracheopexy for membranous intrusion, and posterior descending aortopexy for left bronchus obstruction.

Introduction

Tracheomalacia (TM), characterized by excessive dynamic collapse of the trachea during expiration, is the most common congenital tracheal malformation, with an incidence of 1 in 2100 children.[1-3] If the trachea and mainstem/segmental bronchi are compromised, TM is classified as tracheobronchomalacia (TBM). In children with TBM, the normal C-shaped tracheal cartilages are often replaced with cartilage having a wider and more mobile posterior membrane,[2] an anatomic anomaly which may lead to expiratory collapse. In addition, dynamic airway collapse may occur as a result of external static airway compression by blood vessels or other intrathoracic structures.

The clinical presentation of TBM ranges from expiratory stridor with a typical "barking cough," rhonchi, exercise intolerance, prolonged/recurrent pneumonia, respiratory distress episodes, cyanosis and bronchiectasis to ventilation dependency, apneic events with dying spells, and failure to thrive.[4] In the presence of any of these symptoms, TM/TBM must be considered and investigated.[1,2,4,5] Diagnosis may be difficult since the clinical features are non-specific and some patients are asymptomatic at birth — symptoms may not appear until 2−3 months of age or later.

Clinical management is the best approach for patients with mild or moderate TM/TBM, since malacia may improve as the child grows. However, severe TM/TBM carries a higher risk of complications and life-threatening symptoms; children with severe TM/TBM tend to fail clinical management and usually need surgery.[1,2,6] Defining the best alternative among the available options, which include tracheotomy, tracheal resection, sliding tracheoplasty, anterior/posterior aortopexy, tracheopexy, and stents,[7,8] entails knowledge regarding the association of malacia with external compression, with three-phase dynamic bronchoscopy an essential procedure to determine the location, classification, and severity of malacia.[1] In the present study, the authors report a case series of 20 children with severe TM/TBM treated surgically at the studied institution. Surgical techniques and the outcomes of surgery are described.

Methods

A retrospective study was performed at Hospital de Clínicas of Porto Alegre (HCPA), Brazil. The study was approved by the Institutional Review Board (IRB) and Ethics Committee at HCPA (no. 51093421.8.0000.5327).

A review of medical records of all pediatric patients receiving surgical treatment for severe TM/TBM from January 2003 to April 2023 was performed. Patients were followed until their last recorded visit to the outpatient clinic or emergency room. Data were collected on symptoms, associated comorbidities, previous surgery, age at diagnosis, complementary tests, age at surgery, operative approach, time of follow-up, and outcome at the last follow-up. All the surgical procedures described were done by open surgery and performed or supervised by the same pediatric surgeon (JCF). All patients with available records were included in the study following informed consent by caregivers. Patients were divided into two groups, based on the primary outcome defined as surgical success — symptom improvement.

Data were recorded on Excel® and basic statistical analysis was performed on IBM SPSS Statistics® v26. A normality test was performed for quantitative data and the Mann-Whitney test was used for comparing medians. Fisher's exact test was used to determine associations between qualitative data. Significance was defined as a probability value < 0.05.

Surgical techniques

Bronchoscopy

All patients underwent flexible and/or rigid bronchoscopy pre, intra and postoperatively. A 2.8 mm flexible scope was used for neonatal cases, and a 3.6 mm flexible scope was used for infants and children. Rigid bronchoscopy was performed with a 0° 2.7 or 4 mm scope. For a complete assessment of the structure and function of the airway, 3-phase dynamic bronchoscopy was performed.[1] All endoscopic procedures were performed in the operating room under general anesthesia with spontaneous and vigorous breathing or cough.[1,2] Intraoperative examination was performed through the endotracheal tube to confirm tracheal lumen opening during the surgery.

Anterior aortopexy (AA)

AA was the procedure of choice for patients with malacia and anterior airway compression from the great vessels. The patient was positioned in supine decubitus for partial sternotomy or anterior thoracotomy in the third intercostal space. The thymus was partially or completely resected. For AA, non-absorbable 4-0 or 5-0 Prolene® pledgeted sutures were passed into the adventitia aortic layer and anchored through the full thickness of the sternum.[2] The ascending aorta and/or innominate artery were anchored anteriorly depending on which vessel was causing tracheal compression (as shown in the video_Supplementary Material).

Posterior descending aortopexy (PDA)

PDA was the procedure of choice for patients with left mainstem bronchomalacia or compression. The patient was positioned in a right lateral decubitus. A left posterior thoracotomy muscle-sparing incision was made via the fourth intercostal space. PDA was performed by passing non-absorbable 4-0 or 5-0 Prolene® pledgeted sutures into the adventitia layer on the posterior wall of the descending aorta and anchoring the sutures to the periosteum of the

thoracic vertebra.[9] The goal was to move the aorta laterally and relieve compression on the left main bronchi.

Posterior tracheopexy (PT)

PT was the procedure of choice for patients with tracheal obstruction caused by anterior protrusion of the tracheal membranous wall. The patient was positioned in a left lateral decubitus. A right posterior thoracotomy muscle-sparing incision was made in the fourth intercostal space. The esophagus was dissected, repaired, and moved laterally. For the tracheopexy, 4-0 and/or 5-0 non-absorbable Prolene® pledgeted sutures were passed through the posterior membranous portion of the trachea and anchored to the anterior longitudinal ligament of the spine.[10] Intraoperative bronchoscopy was crucial to confirm tracheal opening and also to make sure the suture did not cross the full thickness of the tracheal posterior membrane.

Combined procedures

In children with more than one etiology of TBM (e.g., anterior compression combined with posterior tracheal intrusion), more than one type of vascular or tracheobronchial pexy can be done.

Results

Characteristics of the sample

Twenty patients were included in the present study and underwent one or more procedures (AA, PDA, or PT) (three patients with two procedures). Eleven patients (55 %) were male and nine (45 %) were female. The median age at diagnosis was 2.5 months (range: 21 days to 12 years). Symptoms at presentation included stridor (n = 10; 50 %), cyanosis (n = 10; 50 %), recurrent respiratory infection (n = 9; 45 %), extubation failure (n = 6; 30 %), dying spells (n = 4; 20 %), apneic events (n = 3; 15 %), atelectasis (n = 1; 5 %), and/or failure to thrive (n = 3; 15 %). The most common underlying conditions were esophageal atresia (n = 8; 40 %), prematurity (n = 7; 35 %), laryngomalacia (n = 5; 25 %), and great vessel anomalies (n = 3; 15 %).

Fourteen patients (70 %) had already undergone surgery, mostly esophageal atresia repair (n = 8; 40 %) and tracheotomy (n = 8; 40 %) due to airway pathologies including micrognathia, laryngomalacia, pharyngomalacia, subglottic stenosis vocal cord paralysis, and macroglossia secondary to Beckwith-Wiedemann syndrome. Other previous procedures included tracheoplasty (n = 1; 5 %), fundoplication with gastrostomy (n = 1; 5 %), endoscopic supraglottoplasty (n = 1; 5 %), and jaw distraction (n = 1; 5 %).

Surgical treatment of TM/TBM

All patients were submitted to preoperative bronchoscopy. TM was more frequent on distal trachea (n = 12; 60 %), followed by mid-to distal trachea (n = 5; 25 %). Six (30 %) patients also presented left mainstem bronchomalacia, and four required surgical management due to severe bronchi compression and TBM.

Median age at surgery was 5.5 months (range: 1 month to 21 years). Eight patients (40 %) underwent anterior transverse thoracotomy, seven (35 %) partial sternotomies, and five (25 %) posterolateral thoracotomy. AA was performed in 15 (75 %) patients, PDA in four (20 %), and PT in four (20 %). In three children, both AA and PT for severe TM were performed due to the association of anterior compression with protrusion of the tracheal membranous wall.

Surgical outcome

All patients were submitted to intraoperative bronchoscopy to guide vascular and tracheal sutures. No patients died during the procedures or in the early postoperative period. Two patients had immediate postoperative complications (10 %): one with wound infection and another one with phrenic nerve injury requiring diaphragmatic plication.

Of the 20 patients, surgery improved respiratory symptoms and was considered successful in 16 (80 %) after a mean follow-up of 12 months (range: 0 to 72 months). Three patients (15 %) who failed surgery died in the long-term follow-up: two due to recurrent respiratory infection, and one who had remained with a tracheostomy died 5 months after surgery due to heavy bleeding through the tracheostomy – probably resulting from an aortotracheal fistula. The other failed patient (5 %) persisted with tracheostomy due to respiratory issues.

Three of eight patients (37.5 %) with previous tracheotomy were decannulated, two in the long term (3 and 5 years after the procedure) and one 7 months after the procedure. One patient with a previous tracheotomy died. One other persisted with tracheostomy due to respiratory problems. In two patients, tracheotomy was maintained in spite of TM improvement after postoperative bronchoscopy: one with severe laryngomalacia, and one other with obstructive macroglossia associated with Beckwith-Wiedemann syndrome.

When comparing the group who improved after the procedure (80 %) with the group who did not (20 %) (Table 1), "dying spells" at the time of diagnosis were observed in three out of four patients with surgical failure and the association was statistically significant (p = 0.01). Statistical association with surgical failure was not detected for any other variables (Table 1).

Discussion

Many patients with TM have mild symptoms that are often not investigated.[2,11,12] Therefore, despite being the most common congenital malformation of the trachea[1] TM is still underdiagnosed. Tracheobronchial collapse may present as an isolated disorder, but as observed in this series, it may also be associated with one or more underlying conditions. No standardized guidelines are available for the evaluation, diagnosis, or treatment of severe TM/TBM, including grading of symptom severity, criteria for radiographic or endoscopic evaluation, medical treatment, and surgical approach.[13] These issues demonstrate the complexity of this condition and underscore the need for assessment and management in centers with a multidisciplinary team specializing in complex pediatric airway disorders.[14]

The clinical presentation of TBM depends on the location, extent, and severity of the airway collapse.[10,14,15] Signs and symptoms range from expiratory stridor to life-threatening

Table 1 Children with or without TM/TBM symptom improvement following surgical treatment in a single center.

	Surgical success *n* = 16/20 (80 %)	Surgical failure *n* = 4/20 (20 %)	p value
Age of diagnosis (months)	2 (0—18)	7.5 (1—144)	0.249
Most common symptoms			
Stridor	7 (43.75)	3 (75)	0.582
Cyanosis	7 (43.75)	3 (75)	0.582
Recurrent infections	8 (50)	1 (25)	0.591
Failure of extubation	5 (31.25)	1 (25)	1.000
Dying spells	1 (6.25)	3 (75)	*0.013*
Common associated conditions			
EA/TEF	7 (43.75)	1 (25)	0.543
Prematurity	5 (31.25)	2 (50)	0.587
Laryngomalacia	3 (18.75)	2 (50)	0.516
Previous surgery			
EA/TEF repair	*7 (43.75)*	*1 (25)*	0.676
Tracheotomy	*6 (37.5)*	*2 (50)*	
Site of malacia			
Proximal to mid-thirds	2 (12.5)	0 (0)	0.637
Mid-third	1 (6.25)	0 (0)	
Mid- to distal thirds	3 (18.75)	2 (50)	
Distal third	10 (62.5)	2 (50)	
Associated bronchomalacia	6 (37.5)	0 (0)	0.267
Age at surgery (months)	4.5 (1—240)	17.5 (3—144)	0.178
Type of incision			
Anterior thoracotomy	5 (31.25)	3 (75)	0.387
Partial sternotomy	6 (37.5)	1 (25)	
Posterolateral thoracotomy	5 (31.25)	0 (0)	
Type of plicature			
AA	11 (68.75)	4 (100)	0.624
PDA	4 (25)	0 (0)	
PT	4 (25)	0 (0)	
AA + PT	3 (18.75)	0 (0)	
Postoperative complications			
Phrenic nerve injury	1 (6.25)	0 (0)	1.000
Wound Infection	1 (6.25)	0 (0)	
Time of follow-up (months)	12 (0—72)	8 (12—18)	0.494
Decannulation (*n* = 8 patients with tracheotomy)	3 (100)	0 (0)	0.066

EA/TEF, esophageal atresia with tracheoesophageal fistula; AA, anterior aortopexy; PDA, posterior descending aortopexy; PT, Posterior tracheopexy.

events. In a literature review,[16] of the most frequent symptoms were acute life-threatening events (43 %), stridor (26 %), and recurrent pneumonia (21 %), while in the present study, the most frequent symptoms were stridor and cyanosis followed by recurrent infections. Diagnosis is sometimes possible in the late neonatal period, although important confounding factors such as prematurity, pulmonary dysplasia, and prolonged mechanical ventilation may hinder recognition.[2]

Dynamic bronchoscopy remains the most important step in TM/TBM investigation, not only to determine the severity, site, and extension but also to evaluate other airway malformations. Endoscopic evaluation should be done in children at high risk of dynamic and/or static airway collapse based on clinical history and examination.[15] Bronchoscopy is performed under general anesthesia, with static, dynamic, and positive pressure evaluations. It is imperative to examine the tracheal rings and to determine the location of malacia and the presence of vascular compression. A lumen reduction greater than 50 % confirms the diagnosis of TM/TBM.[2]

Computed tomography (CT) scans are not useful for TM/TBM diagnosis, since the images may underestimate the severity of the malacia.[7] However, reconstructive CT is advantageous for surgical planning and evaluation of mediastinal anatomy, especially the position of vascular structures such as the aorta in relation to the spine or sternum. CT is also important for the identification of the artery of Adamkiewicz and to guide the surgeon to avoid injuring this critical artery during PDA and mediastinal dissection.[3]

Children with a history of apneic episodes should be also evaluated for cardiac and neurologic causes. In patients with a history of esophageal atresia/esophageal fistula, it is important to evaluate gastroesophageal reflux, esophageal stricture, and recurrent tracheoesophageal fistula, all of which involve symptoms that resemble those of TM/TBM.[2]

Mild and moderate TM/TBM usually do not require surgery, with clinical management being enough to improve symptoms. Furthermore, symptoms may improve as the child grows and the airway enlarges.[1,2,5,6] Conversely, patients with severe symptoms and tracheal lumen obstruction greater than 75 % are at high risk of complications with life-threatening symptoms, thus justifying the need for operative management.[12,17]

The lack of standard classification and management schemes complicates the choice of operative procedure in patients with TM/TBM, and therefore patients need to be evaluated individually.[10,14] There are numerous surgical options for treating severe TM/TBM.[2] It should be noted that tracheotomy is just a temporary treatment since it does not correct TM/TBM directly. Also, tracheal stents have been associated with complications such as stent displacement, perforations, and difficulty or impossibility of removal.[18,19] There is little in the literature about absorbable stents, and more studies are necessary for adequate evaluation.[20] Tracheal resection with primary anastomosis may be useful in cases in which the malacia involves a short segment. In children, it is possible to safely resect one to six tracheal rings. Releasing the larynx and main bronchi during dissection decreases the anastomosis tension and therefore the risk of dehiscence.[21]

The first AA was performed in children in 1948.[3,12] Since then, it has proven effective in the management of some types of severe TM, although the associated morbidity and mortality still raise concern.[1,2,16,17] Complications related to the procedure are bleeding, pleural or pericardial effusion, chylothorax, laryngeal nerve injury, and phrenic nerve injury,[18,22] of which only phrenic nerve injury occurred in the present sample. AA addresses the anterior vascular compression component by indirectly elevating the anterior tracheal wall of the trachea. Based on endoscopic evaluation, the authors believe that AA is not the best procedure to address dynamic posterior membranous airway intrusion. For that, PT is preferable, with significant improvements in clinical symptoms and degree of airway collapse on bronchoscopy.[10] Performing a PT in three of the studied patients with previous AA markedly improved the tracheal collapse because the posterior wall was fixed to the spine. In children with both anterior compression and posterior intrusion of the membranous trachea, more than one type of pexy can be done, as long as a time interval is observed between these procedures.[14]

The effectiveness of AA and PT can be limited by left mainstem bronchomalacia in some patients. The distal left mainstem bronchi is compressed between the descending aorta and pulmonary artery. Surgical options for these patients include PDA, descending aortic translocation, and pulmonary artery fixation.[9] PDA was done in four of the patients with TBM and left main bronchi obstruction, and resulted in improvement of symptoms.

Intraoperative bronchoscopy is also controversial during surgery. A literature review showed that 58 % of the surgeons do not consider intraoperative bronchoscopy necessary.[16] However, like other authors,[1,2,22] the authors consider this as a crucial exam to assess suture placement and confirm adequate airway opening during surgery. Intraoperative bronchoscopy was performed in all patients of this series.

The 80 % success rate in the studied patients is in accordance with the 60–80 % rate reported in the literature.[5,10,11,23] The complication rate in our sample was 10 %, which is smaller than previously reported (12 to 16 %),[1,24] and included wound infection and phrenic nerve injury. Both patients ultimately had a good outcome, the former after an antibiotic course and the latter after diaphragmatic plication.

The presence of "dying spells" at diagnosis in this study was associated with surgical failure. This symptom may signal a complex patient with very severe TM/TBM, requiring extra meticulous surgical planning to adequately determine which portion of vessels and/or trachea must be fixated to achieve the best possible surgical outcome.

A possible limitation of the present study is its retrospective nature, based on a chart review, including clinical presentation, pre and postoperative symptoms, bronchoscopy, results, and follow-up. However, TM/TBM is an uncommon disease in children, and it is usually associated with other diseases and previous surgeries, making it unlikely that a high number of patients can be identified, and preventing the design of prospective studies.

Conclusion

Although TM/TBM is the most common malformation of the trachea, it remains an underdiagnosed condition. Patients have variable clinical presentations, but the investigation can be standardized with diagnostic 3-phase bronchoscopy. CT scans are useful for vascular assessment and surgical planning. The surgical approach should be individualized and based on individual etiology, extension, and location of the TM/TBM. The authors prefer AA for anterior compression, PT for membranous intrusion, and PDA for distal left bronchus compression. The presence of dying spells at diagnosis seems to be associated with surgical failure. The present case series provides new epidemiological data and reinforces the feasibility and effectiveness of surgery for TM/TBM, with no deaths directly associated with the procedures.

Informed patient consent

Informed consent was obtained from the patient's caregivers. The study protocol was approved by the Hospital de Clínicas de Porto Alegre (HCPA) Institutional Review Board (no. 51093421.8.0000.5327).

Previous communication

This paper has not been presented and is not based on previous communications.

Conflicts of interest

The authors declare no conflicts of interest.

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Supplementary materials

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