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Surgical Management of Tracheobronchomalacia in Children: Indications, Results and Techniques Based on a Large Case

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ABSTRACT

Introduction: Pediatric tracheobronchomalacia (TBM) may require surgical treatment for a range of symptoms, from chronic barking cough to severe respiratory episodes. However, there is limited consensus on surgical indications and techniques. Moreover, few reports describe the clinical outcome of TBM surgery, which includes anterior aortopexy (AA) and posterior tracheopexy (PT). Therefore, we evaluated indications, outcomes and surgical techniques in a large series of patients operated at our center for TBM.

Methods: Since 2012, surgery was performed in 143 patients: 132 AA, mostly via mini-sternotomy, and 18 PT via thoracoscopy (14 cases robot-assisted). We evaluated surgical indications and outcome, by assessing the following pre- and postoperative parameters: life-threatening episodes, barking cough, stridor, recurrent respiratory infections, reduced exercise tolerance, and dysphagia.

Results: All patients underwent preoperative dynamic endoscopic evaluation and angio-CT. Surgery (AA, PT or both) was suggested according to a combination of clinical and endoscopic findings, after multidisciplinary team evaluation. Complication and hospital stay rate were similar for AA (18.9 %; 8 days) and PT (16.7 %; 8 days). Clinical improvement was observed in 89.1 % after AA and 93.8 % after PT. Complete resolution of symptoms was observed in 54.6 % after AA and 50 % after PT. Seven patients required both AA and PT.

Conclusion: In our large series of pediatric patients with TBM, surgical indications were based on a combination of clinical and endoscopic findings. Multidisciplinary team discussion is fundamental for correct management of these patients. AA and PT are complementary techniques, both effective and safe.

1. Introduction

Tracheobronchomalacia (TBM) is a condition characterized by an increased susceptibility of the airways to collapse [1]. It can be associated with congenital anomalies such as esophageal atresia and tracheoesophageal fistulas (EA/TEF) [1–3]. The term "TBM" encompasses various conditions, including extrinsic vascular airway compression (as observed in cases of aberrant innominate artery compression or vascular rings), diffuse or focal cartilage weakness resulting in dynamic collapse of the large airways, and excessive dynamic movement of the posterior tracheal membrane during forced exhalation [1–3]. Different regions of the airway may be affected: the collapse may be limited to the trachea (tracheomalacia), or involve one or both main bronchi, the latter condition is referred to as tracheobronchomalacia (TBM) [2].

The most common clinical presentations of TBM are barking cough and biphasic or expiratory stridor [2]. Recurrent respiratory tract infections, feeding difficulties (esophageal dilation could cause tracheal compression), increased work of breathing,

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wheezing, cyanosis, and apneic or dying spells (currently called ALTEs and BRUEs) may also be present [1,2].

Management of TBM is based on the severity of symptoms: mild to moderate forms are always approached with a medical treatment first [2,3]. These therapies include hypertonic saline nebulizer treatments, inhaled low-dose corticosteroids, inhaled ipratropium bromide and chest physiotherapy [2,3]. Continuous positive airway pressure and bilevel positive airway pressure are also non-invasive treatments for TBM [2,3].

The role of surgery is still debated. It is generally believed that TBM may improve as a child grows, leading to the common practice of reserving surgical intervention for severe or debilitating cases. However, there is no consensus in the literature regarding the specific indications for surgery in TBM patients.

Another point of contention is the choice of surgical technique. The primary surgical options include anterior aortopexy (AA), posterior tracheopexy (PT), tracheal resection, internal stenting, and external airway splinting [4,5]. Since its introduction, AA has been recognized as an effective treatment for TBM, with success rates reported to exceed 80 % in most case series [5,6]. PT, on the other hand, is a more recent technique initially developed to reduce the risk of tracheoesophageal fistula (TEF) recurrence, but it has also been shown to effectively address intrusion of the pars membranacea [7,8]. Despite these developments, the literature remains limited with regard to reports on the outcomes of surgical treatments for TBM [4,5]. Considering the lack of scientific evidence on all previous issues regarding TBM, we present our experience with surgical treatment of TBM during the last 12 years in a large series of patients, in an attempt to provide some answer to the following questions: which are the surgical indications to TM? Which factors brought us to perform an AA, a PT or both? What is the expected outcome of these surgeries?

2. Methods

We retrospectively reviewed charts of all children treated surgically for TBM at our Institute between 2012 and 2024. We decided to include all children treated with AA or PT in the considered period and to exclude the patients treated with other procedures (tracheal resection, internal stenting), because the latter were a minority of cases with localized malacia or multiple airway conditions associated.

For all patients, surgical indication was discussed by a multidisciplinary team including pediatric surgeons, cardiothoracic surgeons, pulmonologists, ENTs, gastroenterologists, intensivists, anesthesiologists and radiologists.

The presence or absence of the following clinical parameters, stratified for age, was retrieved in each case:

- 0–2 years: ALTEs; persistent or recurrent barking cough; in- or expiratory stridor; increased work of breathing; recurrent and prolonged respiratory infections affecting the lower tract;
- 2–6 years: persistent or recurrent barking cough; recurrent and prolonged respiratory infections of the lower tract (>6/year); poor resistance to play; in- or expiratory stridor, dysphagia or recurrent vomiting;
- >6 years: persistent or recurrent barking cough; exercise intolerance; cough under stress, not accompanied by bronchospasm; recurrent and prolonged respiratory infections affecting the lower tract (>6/year); dysphagia or vomiting and/or gastroesophageal reflux (GER).

Considering clinical symptoms, a score was assigned to the patients, counting one point for each symptom.

The score has been calculated before and after surgery in order to evaluate clinical evolution.

Diagnosis and site of malacia were assessed by dynamic fibrobronchoscopy (FBS) and chest angio CT. All FBS were performed with a lightly sedated and spontaneously ventilating child. Degree of dynamic airway collapse is determined by the percentage of airway that remains open after cough, forced by stimulating the pars membranacea with the instrument. According to the European Respiratory Society classification, TM was endoscopically described as mild (50–75 % reduction), moderate (75–90 % reduction) or severe (>90 % reduction) based on FBS findings [9]. FBS was always repeated also intra-operatively to help the surgeon and months post-operatively to assess the result.

Data were sourced from case notes, operative records and intensive care information, and included: demographics; comorbidities; diagnosis; surgical approach; operative details; follow-up details. Outcome measures included: post-operative days of hospitalization; complications and their management; clinical and endoscopic outcome. Descriptive statistics were performed with categorical variables reported as frequencies with percentages. AA and PT populations were compared using Fischer's exact test, expressing data as median and interquartile range. All statistical tests were two-sided and P values < 0.05 were considered significant.

Surgery was proposed for symptomatic patients with a mild to severe form of TM documented at FBS, after multidisciplinary evaluation and discussion within the Airway team. All patients treated surgically had received a conservative treatment before, without clinical improvement.

AA was the procedure of choice for symptomatic patients with anterior malacia and/or vascular compression. The procedure was conducted via anterolateral thoracotomy or via mini-sternotomy. The thymus was totally resected in all cases to obtain more space anteriorly. Non-absorbable 4–0 Prolene® stitches were placed between the adventitia aortic layer and the sternum, in infants and small children through the full thickness of the bone. All stitches are pledgeted with pericardial tissue.

PT was the procedure of choice for patients with tracheal occlusion determined by the intrusion of the pars membranacea into the lumen. Most of these patients were treated with a thoracoscopic robotic approach: the patient was positioned in a left lateral decubitus, trocars were placed in line as shown in Fig. 1. Low flow carbon dioxide insufflations was performed to provide adequate visualization, with a pressure of 6 mmHg and a flow that varied from 2 to 4 L/min. The first step of PT was azygous vein ligation and division.

Then mediastinal pleura was opened longitudinally and the vagus nerves, the esophagus, the airway were identified. After esophageal dissection, the esophagus was encircled by a silastic loop and then retracted laterally to ease the access to the trachea. Identification of the bulging pars membranacea was helped by intraoperative ventilation and better identified with bronchoscopy, performed inserting a small bronchoscope into the tracheal tube. The anterior body of the spine was prepared, and anterior spinal ligament was identified. Multiple non-absorbable stitches of 4–0 Prolene® were placed transversally between the pars membranacea and the anterior spinal ligament, avoiding entering the airway lumen (checked bronchoscopically) as shown in Fig. 2. Thoracic drain was left in all cases.

3. Results

During the study period, 143 patients underwent surgery for TBM. The characteristics of the study cohort, stratified by age, are summarized in Table 1. Main etiology of TBM was extrinsic anterior

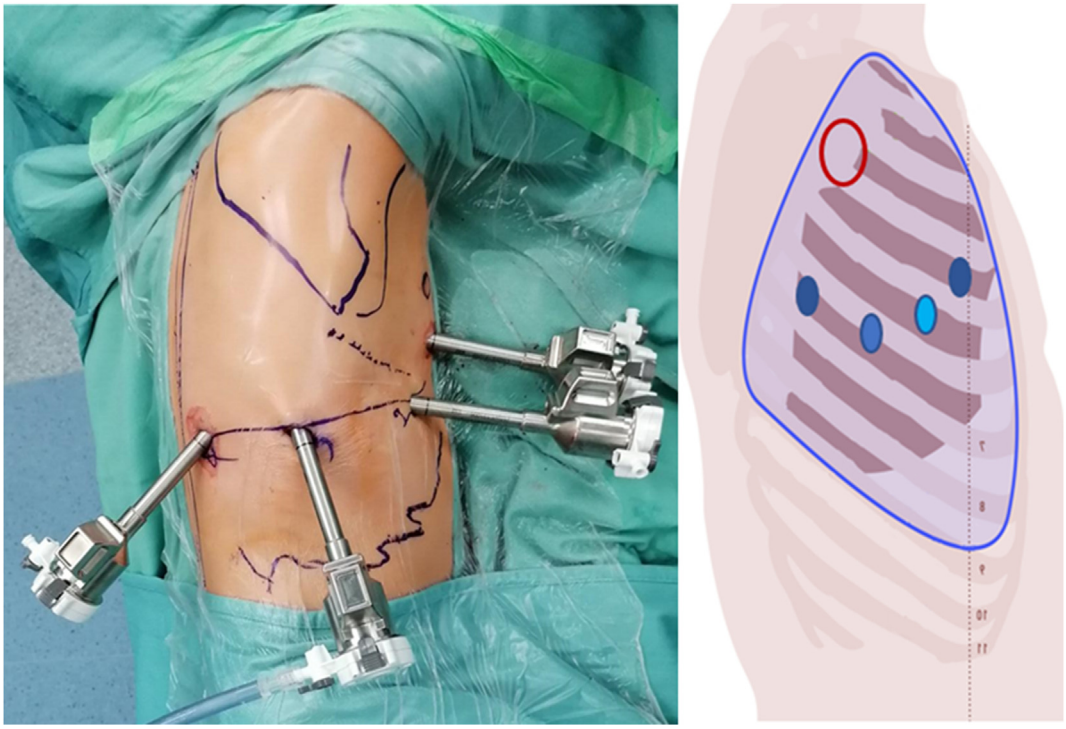

Fig. 1. Trocars placement for robot-assisted thoracoscopic posterior tracheopexy

innominate artery compression and subsequently the majority of patients presented anterior TBM and underwent AA (Table 1).

Of the 143 patients, 132 underwent AA, 18 underwent PT, and 7 patients received both procedures (6 of these in two separate operations and 1 in a single combined procedure). Surgical details are provided in Table 2. Anterior ministernotomy was the approach used for AA in 108 patients (81.8 %), while the remaining 24 patients underwent a lateral thoracotomy. PT was performed using a video-assisted thoracoscopic surgery (VATS) approach in 4 patients, until 2020, and a robotic-assisted thoracoscopic surgery (RATS) approach in the subsequent 14 patients. In patients treated with VATS approach, the number of stitches placed in surgery was 3 ± 2 (range: 2–6) during an average operative time of 180.5 ± 94.6 min (range: 110–320). In patients treated with RATS approach, the number of stitches placed in surgery was 8 ± 2 (range: 6–14) and the console operative time 129.8 ± 47.4 min (range: 75–215). In specific cases of TM associated with bronchomalacia, robotic assistance allowed the placement of one or two stitches on the main bronchi addressing the problem.

Follow-up clinical data were available for 110 of the 132 AA patients and 16 of the 18 PT patients, while endoscopic data were available for 98 AA patients and 13 PT patients. The mean follow-up period was 29 ± 25.3 months (range: 5.2–130.7) for patients who underwent AA and 9.8 ± 11.2 months (range: 1.4–40.6) for those

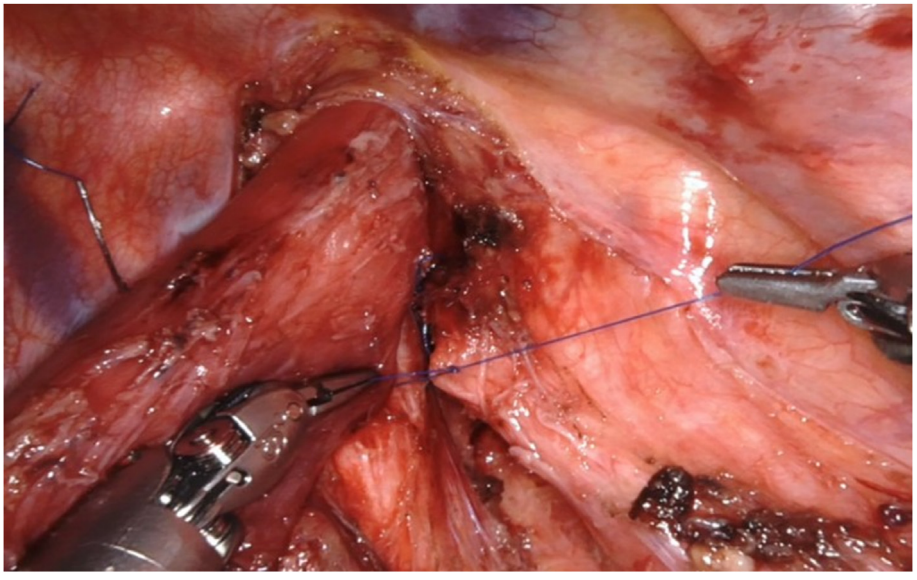

Fig. 2. Suture between the anterior spinal ligament and the pars membranacea.

Table 1
Patients characteristics stratified by age.

Characteristics	Total Cohort (N = 143)	0–2 years (N = 43)	2–6 years (N = 63)	≥6 years (N = 37)
	n (%) or median±SD (IQR)	n (%) or median±SD (IQR)	n (%) or median±SD (IQR)	n (%) or median±SD (IQR)
Demographic				
Male	88 (61.5 %)	28 (65.1 %)	39 (61.9 %)	21 (56.8 %)
Age at 1stsurgery (months)	68 ± 56 (2–278)			
Patient history				
EA/TEF	41 (28.7 %)	22 (51.2 %)	13 (20.6 %)	6 (16.2 %)
CV malformation	12 (8.4 %)	7 (16.3 %)	5 (7.9 %)	–
Other comorbidities	35 (24.5 %)	19 (44.2 %)	13 (20.6 %)	3 (8.1 %)
CPAP/MV	13 (9.1 %)	8 (18.6 %)	1 (1.6 %)	4 (10.8 %)
Clinical score				
1 point	34 (23.8 %)	13 (30.2 %)	13 (20.6 %)	8 (21.6 %)
2 points	83 (58 %)	23 (53.5 %)	38 (60.3 %)	22 (59.5 %)
3 points	25 (17.5 %)	7 (16.3 %)	12 (19.1 %)	6 (16.2 %)
4 points	1 (0.7 %)	–	–	1 (2.7 %)

TM = tracheomalacia, EA/TEF = esophageal atresia/tracheoesophageal fistula, CV = cardiovascular, CPAP=Continuous positive airway pressure, MV = mechanical ventilation.

Table 2
Details of surgical procedures.

Parameters	AA (N = 132)	PT (N = 18)	*P value*
	n (%) or median±SD (IQR)	n (%) or median±SD (IQR)	n
Weight at surgery (Kg)	17 ± 15 (4–60)	51 ± 21 (15–67)	<0.001
Age at surgery (months)	55 ± 49 (2–200)	125 ± 68 (41–279)	<0.001
Hospital stay (days)	8 ± 7 (4–50)	8 ± 11 (3–16)	0.4956
Complications			
• Overall	25 (18.9 %)	3 (16.7 %)	>0.99
• Grade ≥3	3 (2.3 %)	1 (5.6 %)	0.4036

who underwent PT. Complications were infrequent in both populations with a similar overall complications rate *($p > 0.99$)* and a similar rate of complications which required subsequent intervention ($p = 0.4036$); frequences are reported in Table 2. For AA, we observed a case of intra-operative aortic laceration while the main post-operative complication was pericardic effusion treated mostly conservatively (one case only required subsequent drainage). In PT population, no intra-operative complication was observed, while post-operative complications consisted in thoracic duct lesion in 2 patients (conservative treatment) and esophageal perforation in 1 patient previously operated for EA/TEF, treated with a stent positioning.

Considering endoscopic outcome, an improvement, determined as a downgrade of the endoscopic score, was observed in 87.8 % of AA patients and 92.3 % of PT patients ($p > 0.99$). Clinical improvement, determined as a downgrade of the clinical score, was achieved in 89.1 % of AA patients and 93.8 % ($p > 0.99$), while the complete resolution of symptoms was observed in 54.6 % vs 50 % respectively ($p = 0.7924$). The comparison between original and follow-up clinical scores is reported in Table 3 and a statistically significant difference has been found between pre- and post-operative for almost all scores. Regarding the 13 patients who were treated pre-operatively with continuous positive airway pressure (CPAP) or mechanical ventilation: 8/13 (61.5 %) stopped this treatment after surgery due to clinical improvement, 2/13 (15.4 %) continued it and 3/13 (23.1 %) were lost at follow-up.

4. Discussion

TBM is a relatively common condition, with an estimated incidence of 1 in 2100 in the European population [10], potentially leading to significant morbidity in children [9]. Although some believe that children, particularly those without associated syndromes, may outgrow the condition, there are limited data to accurately predict which children will improve and which will not [9]. There is a general lack of consensus on surgical management and indications for TBM. Despite the relatively common incidence of TBM and its potential severity, only few papers in the literature report large series of patients treated surgically for TBM. In 2024, Sutton et al. [5] reported on a large cohort of 169 patients treated with open and thoracoscopic AA for TBM. They demonstrated that AA is a safe and effective treatment, with satisfactory successful extubation, tracheostomy decannulation, and survival rate. However, the Authors did not assess symptoms or endoscopic outcomes and studied only AA.

Shortly after, Mukaresh et al. [4] published the outcome of 73 TBM patients treated with various surgical techniques, including AA, PT, and splinting. This study, which was the first to consider all these techniques, provided detailed clinical and endoscopic outcomes. However, the series was limited to a cohort with primary TBM, excluding secondary cases. To our knowledge, our study, though with the bias of a retrospective analysis, reports the largest series in the literature of surgically treated TBM pediatric patients, in which symptoms, intervention types and outcomes were studied.

TBM patients are usually referred to our Airway team by the pulmonologists, after an unsuccessful period of medical treatment. We advocate a multidisciplinary team approach in order to evaluate for every patient the symptoms, the previous medical treatment, the endoscopic and radiological findings and the comorbidities. Surgical indications were discussed at the multidisciplinary team taking into account all previous data. In our experience, it is not always easy to decide which patients could benefit from surgery. Endoscopic and clinical presentation may not be always corresponding. The combination of clinical and endoscopic findings (and not only one of them) should be considered when indicating surgery for TBM. In an effort to have an objective tool to evaluate the degree of severity of TBM in our patients, we adopted a clinical score based on symptoms and an endoscopic classification of TBM, proposed by ERS.

Table 3
Comparison between pre and post-operative clinical score.

	Pre-op Cohort (N = 143)	Follow-up Population (N = 116)	*P value*
	n (%) or median±SD (IQR)	n (%) or median±SD (IQR)	*n*
Clinical score			
0 points	0 (0 %)	68 (58.6 %)	$p < 0.001$
1 point	34 (23.8 %)	47 (40.5 %)	$p = 0.0046$
2 points	83 (58 %)	10 (8.6 %)	$p < 0.001$
3 points	25 (17.5 %)	1 (0.9 %)	$p < 0.001$
4 points	1 (0.7 %)	0 (0 %)	$p > 0.99$

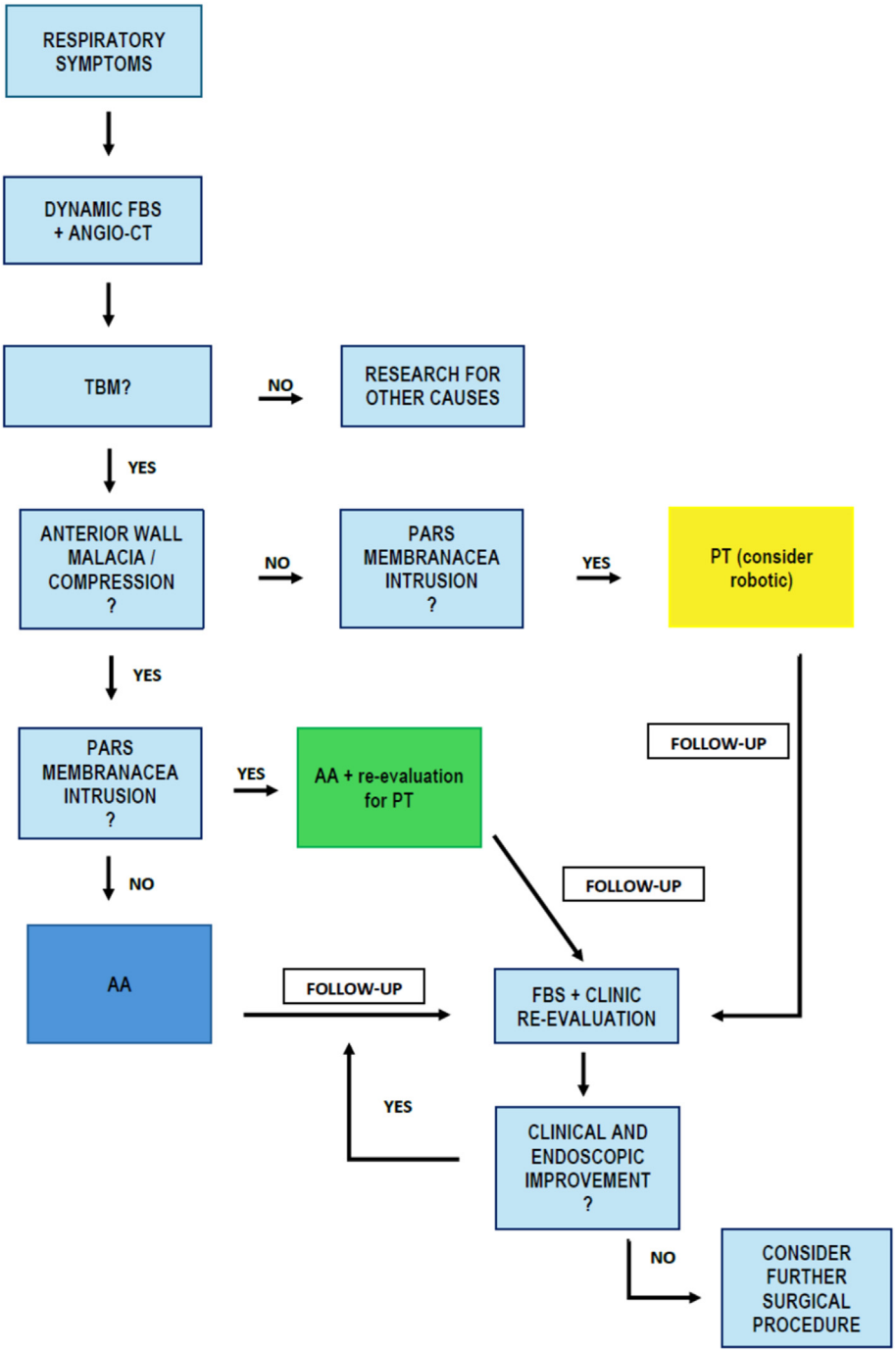

Fig. 3. Algorithm proposed for management of tracheomalacia.

Surgery was considered in patients with mild to severe TBM at dynamic endoscopic evaluation, associated to respiratory symptoms which had a significant impact on the patients' life (for example leading to multiple hospitalizations or poor quality of life). The clinical score that was adopted retrospectively in the present study to evaluate objectively the symptoms of the patients before and after the operation, could be in the future a tool to help the multidisciplinary team to indicate a surgical approach, based on an objective parameter.

A key point made by Mukaresh et al. [4] in their recent paper is that since TBM rarely occurs in isolation, a standardized approach is inadequate; the treatment must be customized to address associated comorbidities and the unique features of each patient. In recent years, our team has become a referral center for diagnosing and managing TBM. We strongly agree with the vision of a custom made approached tailored on the patient. However, we believe that there are some principles which should be considered to direct the treatment in one direction rather than another. The endoscopic findings are essential in order to indicate the proper surgical approach. First of all, our experience confirmed what reported in the literature, AA is an effective and safe treatment for the majority of cases of TBM [4,5]. In the past, a lateral thoracotomy has been performed for AA and in literature are reported several case series of thoracoscopic approach; however, in our experience, the mini-sternotomy has proved to be the better option, with a fast, precise and low invasive approach to the aorta. Therefore, it became the standard for all patients subjected to AA, regardless the age. The mini-sternotomy approach offers also the possibility to perform the thymectomy, a surgical step which ensure further space to the trachea. As AA widens the tracheal lumen by pulling the anterior wall of the trachea together with the aorta, it is an effective approach when TBM is due to anomalous cartilaginous rings or to an extrinsic compression. These two situations considered together represent a great number of cases of TBM, corresponding to the majority in our experience. This explains why we have performed much more AA than PT, not only historically (PT was introduced only 2020 in our Center), but also in the last 5 years. Our results demonstrate that the majority of patients treated with AA (89 %) achieved a satisfactory clinical and endoscopic outcome, not requiring additional procedures. PT, on the other hand, addresses the cases of TBM related to an intrusion of the pars membranacea occluding a significant part of the lumen. It is a relatively recent technique compared to AA, but it has proven to be effective and safe [4,7,8] with the right indications. In our series, hospital stay, complications rate, clinical and endoscopic outcomes are all comparable between AA and PT. The main difference we found was about the age and weight at surgery. In our opinion, this is related to the different indications for the two techniques and also to the recent introduction of PT.

According to our series, some patients treated firstly with AA required later a PT. This can occur when both an anterior collapse of cartilage rings and a posterior intrusion are observed at dynamic endoscopic evaluation. In these patients, AA alone can be not completely sufficient to improve tracheal lumen. It can be debatable if AA and PT can be performed at the same time, which could be theoretically an attractive option. We have performed AA and PT during the same operative session only once. We prefer to perform AA and PT in two steps, as the two procedures require completely different approaches: mini-sternotomy for AA, right thoracoscopy for PT.

We prefer to perform AA and PT in two steps also because it is difficult to predict the need of PT after AA: in some patients we observed a clinical improvement after AA which allowed us to avoid subsequent PT. In Fig. 3 we propose an algorithm which summarizes our approach for the surgical management of TBM.

Alternative surgical techniques for TBM include tracheal resection, stenting and splint application. Tracheal resection may be considered for highly selected patients with very severe short-segment malacia, when other surgical or endoscopic techniques have failed. The complications of a resection are potentially more severe compared to those of AA and PT [11].

Internal stenting in children is appealing but faces several challenges, making it a less common option. Typically reserved for cases where surgery is not viable and tracheostomy is unsuitable, stenting may provide temporary improvement. However, stents can lead to complications such as mucus encrustation, migration, granulation tissue, or stent fracture [12].

Extraluminal splinting may offer effective airway support in highly selected, very severe and/or diffuse TBM cases [13]. Biocompatible ceramic rings, resorbable plates or even three-dimensional printed biodegradable splints have all been used. Possible erosion into surrounding structures, a strangulation effect after somatic growth in a small child, infection and long-term tissue tolerance are concerns. Few reports have been published till today and this treatment is at its very beginning [9,13].

In conclusion, TBM is a pediatric condition requiring a multidisciplinary and patient tailored approach. AA and PT are both effective and safe procedures which can be indicated for treatment of severe forms of TBM. Indications have to be widely discussed within a multidisciplinary team and based on clinical and endoscopic evaluations.

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