

## **Thoracoscopic aortopexy for symptomatic tracheobronchomalacia**

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## Abstract

**Aim:** Symptomatic tracheobronchomalacia can be fatal. Successful treatment includes aortopexy. We report outcomes of the thoracoscopic approach in a single centre.

**Methods:** All patients undergoing thoracoscopic aortopexies from 2009 to 2018 were retrospectively reviewed. Data was reported as median (interquartile range). Risk factors for subsequent tracheostomy were analysed with logistics regression model, P-value<0.05 as significant.

**Results:** Twenty-one patients underwent thoroscopic aortopexy with a follow up duration was 365 (72 – 854) days mid to distal tracheomalacia (n=17) and bronchial involvement (n=4) as defined by bronchoscopy, bronchogram or CT thorax y . Preoperative patient demographics and comorbidities e.g. gastro-oesophageal reflux disease, prematurity and cardiac anomalies were recorded. Indications for thoracoscopic aortopexy were apparent life-threatening event(s) (n=14), recurrent chest infections (n=5) and failure to wean invasive ventilation (n=2). Thoracoscopic aortopexies (n=20) with conversion to open (n=1) were performed. Intraoperative bleeding (n=2) occurred, and chest tube (n=1) was inserted for monitoring. Intraoperative bronchoscopy (n=17) confirmed improvement of tracheomalacia. Anaesthetic time was 140 (90 - 160) minutes. Postoperatively, 2 patients had dehiscence of the aorta from the sternum and underwent revision surgery with posterior tracheopexy. Three patients required tracheostomies. Potential risk factors for subsequent tracheostomy were investigated: only the association of tracheobronchomalacia was close to significance (OR 16 (95% CI 0.95 -267.03), P=0.05).. Symptoms resolution occurred in n=17 (81%) of patients.

**Conclusion:** Different modalities were used to delineate the site of tracheobronchomalacia and its aetiology. Tracheomalacia with bronchial involvement may be a risk factor for subsequent tracheostomy.

**Keywords:** tracheobronchomalacia, esophageal atresia, thoracoscopic, aortopexy, tracheostomy, bronchoscopy, tracheobronchogram

**Level of evidence:** IV

## Introduction

Tracheobronchomalacia (TBM) is a poorly defined however it is typically used to describe a weakness in the trachea and bronchus/bronchi. Baxter and Dunbar[1] defined tracheomalacia (TM) as a “condition in which there is weakness of the tracheal wall due to softening of the supporting cartilage and hypotonia of the myoelastic elements”. It has been further classified as primary (congenital) and secondary (acquired), and now used to describe: (i) unilateral airway compression due to innominate artery or aberrant subclavian artery compression; (ii) circumferential airway compression due to double aortic arch or vascular ring; (iii) diffuse or focal cartilage weakness with dynamic collapse of the large airways (as seen in certain genetic conditions and tracheal inflammation); or (iv) excessive dynamic movement of the posterior tracheal membrane during forced exhalation[2].

TBM can occur in isolation or secondary to the above conditions, affecting variable regions of the airway. It can also be classified to these sites: tracheomalacia (anteroposterior collapse of the whole or segments of the trachea, especially on coughing or expiration); bronchomalacia, a less common entity describing isolated collapse of one or both main stem bronchi[3]; and small airway malacia, involving the lobar bronchi and smaller airways, typically associated with prematurity and bronchopulmonary dysplasia[4].

Aortopexy has been long established, first described by Gross[5] in 1948, where early studies have shown it to be effective in children[3, 6]. Several approaches have been described including, sternotomy (partial[7] or full[3, 6]), thoracotomy[7], suprasternal[8] and thoracoscopic[9], the latter arising recently as a result of improved thoracoscopic equipment and techniques. Currently the outcomes on minimally invasive techniques have been limited, however the hope has been these techniques confer benefits such as less postoperative pain, faster time to feeds and shorter intensive care unit (ICU) and hospital stay. This report describes the characteristics and outcome of the thoracoscopic aortopexies in selected children with symptomatic tracheobronchomalacia from a single unit centre.

## Methods

This was a retrospective consecutive case series of children undergoing thoracoscopic aortopexy for symptomatic tracheobronchomalacia over a 10 years period from 2009 – 2019 at Great Ormond Street Hospital, NHS Foundation Trust, United Kingdom. The study was registered with the institutional clinical audit department, no further ethical approval was required.

All patients had undergone a personalized workup which included direct endoscopy, either microlaryngobronchoscopy (MLB) or flexible bronchoscopy, contrast computed tomography (CT) thorax, and tracheobronchogram to determine the level of malacia. All patients were evaluated through the Aerodigestive MDT. The decision for surgery and the approach used was based upon the investigations above.

Case notes, operative and intensive care data were reviewed and recorded in a registered database. Data were collected on patient gender, age, gestation age, weight, comorbidities, cardiovascular anomalies, gastro-oesophageal reflux disease, previous TOF repair, preoperative endoscopy, CT scan and tracheobronchogram. Perioperative variables recorded were total anaesthetic time, median intraoperative end tidal CO<sub>2</sub>; time to: extubation, off supplemental oxygen, off morphine, first feeds; duration of: ICU stay, hospital stay, follow-up; recurrence of symptoms, scoliosis or chest wall deformities, redo aortopexy and tracheopexy. Primary outcome was subsequent tracheostomy, indicating failure of the aortopexy.

Major cardiovascular anomalies were defined as either cyanotic congenital heart disease that required palliative or corrective surgery, or non-cyanotic congenital heart disease that required medical or surgical treatment for cardiac failure. The site of malacia was classified from direct endoscopy, tracheobronchography or CT scan into TM or TBM. Malacia was defined as collapse of the anteroposterior diameter of the trachea of more than 50%, associated with clinical symptoms.

Data are presented as median with interquartile range (IQR) and/or binomial percentages where appropriate. Logistic regression analysis using Stata 14 SE (StataCorp, Texas, USA) was used to investigate possible risk factors for failure of aortopexy i.e. subsequent tracheostomy. A P-value of less than 0.05 was taken as statistical significance.

## Technique

After endotracheal tube intubation, the patient was placed in a supine position with the left side slightly elevated. A left thoracoscopic approach (Figure 1) avoided the adhesions from

previous right-sided operation e.g. for oesophageal atresia repair. A 5 mm Hassan port was placed at the anterior axillary line at the nipple level and a 5 mm blue port and 3 mm port were placed just anterior to the 5 mm port on each side. After gaining entry into the thorax, the left lobe of the thymus was excised giving good visualisation of the pericardium and aortic arch. The first 3-0 pledgeted polypropylene (Prolene®) suture was passed via a skin stab incision through the sternum, through the adventitia and pericardium overlying the proximal aortic arch. This suture was then fed back through the sternum using an Endoclose®. This technique was repeated with the second and third sutures placed sequentially through the proximal aorta and junction of the aorta with the heart. A flexible bronchoscopy was used to visualise the site of the malacia, and under direct vision, the sutures were held taut (Figure 2&3). When there was significant improvement of the calibre of the trachea (Figure 4), the sutures were tied snugly to the sternum. If there was no or minimal change to the tracheal lumen, the decision was made to either put in more sutures proximally or convert to open. Absorbable sutures and skin glue were used to close the stab wounds. The schematic diagrams (Figures 5) shows the thoracic anatomy before and after thoracoscopic aortopexy.

## Results

### Patient Characteristics

Thoracoscopic aortopexy was performed on 21 patients with symptomatic trachea(broncho)malacia (Table 1). Sixteen (76%) patients were born term, 13 (61.9%) patients were male and corrected age was 194 (83 - 277) days old. The median weight was 5.7 (4.3 – 8.1) kg. Their underlying diagnoses were predominantly (76%) oesophageal atresia with trachea-oesophageal fistula (OA-TOF). Four patients had laryngeal cleft, of which there were Type 2 (n=2), Type 3 (n=1) and Type 4 (n=1) clefts. Three had their laryngeal cleft repaired before the aortopexy, while 1 patient with Type 2 laryngeal cleft was repaired after the aortopexy. Seven (33%) patients had associated symptomatic gastro-oesophageal reflux disease (GORD) and underwent a laparoscopic fundoplication and gastrostomy – 5 patients prior to the aortopexy, and the rest after. Fifteen (71%) patients had a normal cardio-echogram, and 1 patient had a corrected major cyanotic congenital heart disease. One patient with a previous OA-TOF repair at a different centre, underwent a subsequent right thoracotomy, excision of residual tracheal cyst, posterior tracheopexy and aortopexy but had persistent symptomatic tracheomalacia. None had vascular rings or pectus excavatum.

### Site of malacia

Seventeen patients had mid to distal isolated tracheomalacia with bronchial involvement in four. Tracheo(broncho)malacia was due to vascular compression from the innominate artery in 9 patients but was indeterminate in 6 due to the endotracheal tube obscuring the trachea.

### Perioperative findings

The indication for surgery was predominantly for apparent life-threatening event(s) (75%) (Table 1). Of the patients who had preoperative NIPPV (n=5), 2 had recurrent apparent life-threatening events (ALTEs) and another 2 progressed to intubation.

Intraoperatively, 2 patients had bleeding when the suture was placed into the aorta. This was stopped with local pressure, and did not require perioperative blood transfusion. Chest tube was placed in 1 of them for monitoring. Recovery was uneventful in both patients and, on the second patient, the chest tube was removed on postoperative day 2. Intraoperative bronchoscopy (n=17) confirmed resolution of tracheomalacia. Thoracoscopic converted to open aortopexy via a limited manubriotomy (n=1) was performed because intraoperative bronchoscopy showed more proximal vascular compression.

Patients had a preoperative haemoglobin of 111 (99.5 – 120) g/L. The overall end tidal CO<sub>2</sub> (EtCO<sub>2</sub>) was 6.0 (5.4 – 6.3) kPa, although the peak EtCO<sub>2</sub> was high at 7.6 (6.2 – 8.2) kPa. The total anaesthetic time was 140 (90 - 160) minutes.

Postoperatively, the duration of invasive ventilation was 16 (13 – 24) hours, with 16 patients being extubated within 24 hours. The time to wean off non-invasive positive pressure ventilation and supplemental oxygen was 7 (0 – 38) and 2 (0 – 20) hours respectively. The median intensive care unit stay was 2.4 (1.7 – 5.5) days. The time to wean off morphine was 24 (17 – 45) hours and time to tolerate full feeds was 32 (22 – 48) hours. The median hospitalisation stay was 39 (4 – 92) days.

### **Outcome**

Total length of follow up was 365 (72 – 854) days. None of the patients had chest wall deformities or cosmetic concerns. However, 4 (19%) patients had recurrent symptoms. Two patients required redo open aortopexy with posterior tracheopexy at postoperative 1.5 month and 3 months respectively. Both were patients with previous OA-TOF repair. The suture had torn through the adventitia, resulting in dehiscence of the aorta from the sternum. One patient could not be weaned off non-invasive ventilation and underwent a subsequent tracheostomy at the local hospital. The other 2 patients underwent a tracheostomy at postoperative 25 days and 30 days respectively. One had a previous Type 3 laryngeal cleft repair and the other had a previous OA-TOF repair. Both had tracheobronchomalacia and were unable to wean off non-invasive ventilation.

The possible risk factors for failure of aortopexy i.e. need for tracheostomy (n=3) were investigated (Table 2) using a logistics regression model. Only the presence of bronchial involvement tended towards a subsequent tracheostomy, although this did not reach statistical significance (P=0.05). All the other variables for example, body weight, prematurity, syndromic or the presence of a laryngeal cleft were not associated with a subsequent tracheostomy.

## Discussion

Tracheobronchomalacia (TBM) is a rare condition with variable definitions. There is limited evidence in optimal treatment regimens and numerous options have been advocated [10], [11]. Non-invasive positive pressure ventilation (NIPPV) has been used to increase the tracheal intraluminal pressure to prevent its coaptation during expiration, however it is often not tolerated, and it is not uncommon for children to have facial flattening (68%) as a result of prolonged use. Dependency on a ventilatory machine carries the added risk of respiratory compromise as a result of mechanical failure [12]. Stenting of the airway is also feasible however in children is not without complications where obstructive granulation is common requiring regular endoscopic treatment. Furthermore, in children fixed size stents do not accommodate for growth seen in the airway [13, 14].

Surgical treatment for malacia includes aortopexy, tracheal resection and anastomosis, sliding tracheoplasty, pulmonary artery pexy, anterior or posterior tracheopexy, tracheobronchopexy or external splinting[2, 3]. Success rates have varied across centres, however aortopexy has remained a time-tested procedure for tracheomalacia. Open surgery has been used in many centres however increasing success with the thoroscopic approach has led to its adoption in a number of centres[7, 9, 15]. The thoroscopic approach has the particular benefit of earlier time to extubation, shorter hospital stay and smaller scars in neonates[16]. In our unit aortopexy has been predominantly via a median sternotomy, however given the benefits described, an increasing number of children have become a candidate for the thoroscopic approach.

Our experience has shown this approach to be effective and comparable to outcomes via open approaches. In our cohort, there was no need for single lung ventilation, and we have found the approach from the left side has meant minimal adhesions encountered This is particularly as the majority of our patients had OA-TOF repairs which were repaired from the right. Interestingly the only patient who had a redo thoroscopic aortopexy had an initial approach via a right thoracotomy which may reflect access for accurate suture placement .

In adopting this approach, due to use of CO<sub>2</sub> gas insufflation for the thoroscopic approach, appropriate anaesthetic monitoring of end tidal CO<sub>2</sub> (EtCO<sub>2</sub>) has been essential due to the risk of hypercapnia and reduced cerebral oxygenation [17, 18] [19, 20]. Whilst patients had normal preoperative haemoglobin levels, the peak EtCO<sub>2</sub> was high, but expediently corrected to achieve a median EtCO<sub>2</sub> within normal range. Close anaesthetic monitoring is required to prevent this and the EtCO<sub>2</sub> was monitored at 15 minutes interval with the insufflation pressure set to as low as possible, typically 5 - 8 mmHg and flow rate of 1 – 2 L/min.

We have also found intraoperative flexible bronchoscopy to be essential in confirming appropriate opening of the airway prior to tying the knots of the pledgeted sutures to the sternum. We have found this to be an important step that significantly contributes to reduced rates of failure and reflected in our reduced rates of revision and tracheotomy. This is in comparison to other units that report recurrent symptoms in 33% on thoracoscopic aortopexies[15]. A common feature is sutures tearing through which may reflect the learning curve and hesitancy to place sutures deep enough into the aortic wall. Accurate suture placement appears to be important in reducing recurrent symptoms where proponents of open surgery suggest it allows for greater exposure of the great vessels and improve ease of suture placement.

We have found patient selection is important in the decision to perform an aortopexy, and only perform it in cases where there is short segment of malacia that is attributable to vascular compression. Currently we offer a thoracoscopic aortopexy when there is no requirement to address a vascular anomaly. If there is concomitant cardiovascular malformation that requires correction a median sternotomy is normally performed. Stenting is typically performed only if the patient is not candidate for an aortopexy or if there is longer segment malacia. In children the preference is to use biodegradable stents. Most reports describing aortopexy describe only anterior fixation, however thoracoscopic aortopexy has been combined with posterior tracheopexy, offering full circumferential tension on the airway [21]. Whilst this is an attractive option to improve airway calibre, currently we do not feel it is necessary in all cases. We believe this adjunct procedure may be used in select cases where posterior malacia is more evident, or if repeated failures are seen with the anterior approach.

In our series the association of tracheostomy with bronchial involvement was close to significance. This finding was also observed in our units previous report describing outcomes from median sternotomy [22]. More extensive malacia may reflect intrinsic weakness in the airway wall. This is notable with laryngeal cleft repair, where the tracheobronchomalacia is often persistent despite aortopexy. This group remains challenging as the cleft often has considerable long segment disruption of the tracheal framework. Other groups [8] have associated failure in more syndromic patients however we saw no such association

This report was a retrospective, single centre review with its inherent recorder bias which is a limitation. The study also reports from a select group of patients deemed suitable for surgery and may lead to a degree of bias. Despite this, we report the largest series of thoracoscopic aortopexy with detailed follow up. Our experience demonstrates how a thoracoscopic aortopexy is a safe alternative to the open approach, leading to resolution in the majority of

patients. We would advocate this as a suitable approach in selected patients where we see improved recovery time and reduced morbidity.

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**Table 1: Preoperative characteristics of patients. IQR: Interquartile range. N: Number of patients.**

<b>Aetiology (N)</b>	Oesophageal Atresia (15) Laryngeal Cleft (3) Unknown (2) Oesophageal atresia and laryngeal cleft (1)
<b>Median corrected age/days (IQR)</b>	194 (83 – 277)
<b>Median weight/kg (IQR)</b>	5.7 (4.3 – 8.1)
<b>Gender (N)</b>	Male (13) Female (8)
<b>Comorbidities (N)</b>	GORD (7) Ex-prematurity (5) Syndromic (4) Bilateral VC palsy (1)
<b>Cardiac Function (N)</b>	Normal (15) VSD (3) ASD (2) Repaired major cardiac anomaly (1)
<b>Site of malacia (N)</b>	Mid to distal trachea (17) Tracheobronchomalacia (4)
<b>Indications for surgery (N)</b>	Apparent life-threatening event(s) (14) Recurrent pneumonia (5) Failure to wean invasive ventilation (2)

**Table 2: Possible risk factors for failure of aortopexy**

<b>Risk factors</b>	<b>Odds Ratio (95% CI)</b>	<b>P-value</b>
Tracheobronchomalacia	16 (0.95 – 267.03)	0.05
Preoperative non-invasive ventilation	10 (0.67 – 149.04)	0.10
GORD	0.1 (0.01 – 1.49)	0.10
Prematurity	7.00 (0.50 – 98.60)	0.15
Preoperative invasive ventilation	8.5 (0.37 – 195.4)	0.18
Weight	0.39 (0.09 – 1.68)	0.21
Age	0.99 (0.98 – 1.01)	0.38
Absent intraoperative bronchoscopy	2.5 (0.17 – 37.26)	0.51
Aetiology	0.4 (0.03 – 5.96)	0.51
Male gender	2.5 (0.17 – 37.26)	0.51
Cardiac anomaly	2 (0.13 – 30.16)	0.62

## Figure Legends

Figure 1: Thoracoscopic view of the mediastinum from the patient's left side. Part of the thymus overlies the aorta and will be excised.

Figure 2: Thoracoscopic view of the aorta with sutures 'hitched' to the sternum with pledgeted polypropylene (Prolene®) sutures that are passed through the vessel's adventitia.

Figure 3 Schematic diagram demonstrating the mode of action with an aortopexy. (A) Preoperatively, the trachea (t) is crescent shaped with collapse in an anteroposterior direction. (B): 3mm instruments are introduced through the left chest. (C-D) Thoracoscopic approach partial removal of the thymus (th) allows for access to the aortic arch (aa). (E) 3 pledgeted polypropylene sutures (blue) are placed through the adventia of aortic arch (at the base of innominate artery) and passed through the sternum. Sutures are tied such it brings aortic arch (aa) anterior and subsequently brings the anterior tracheal wall forward. (F) The result is an increase in the airway caliber (arrows).

Figure 1.

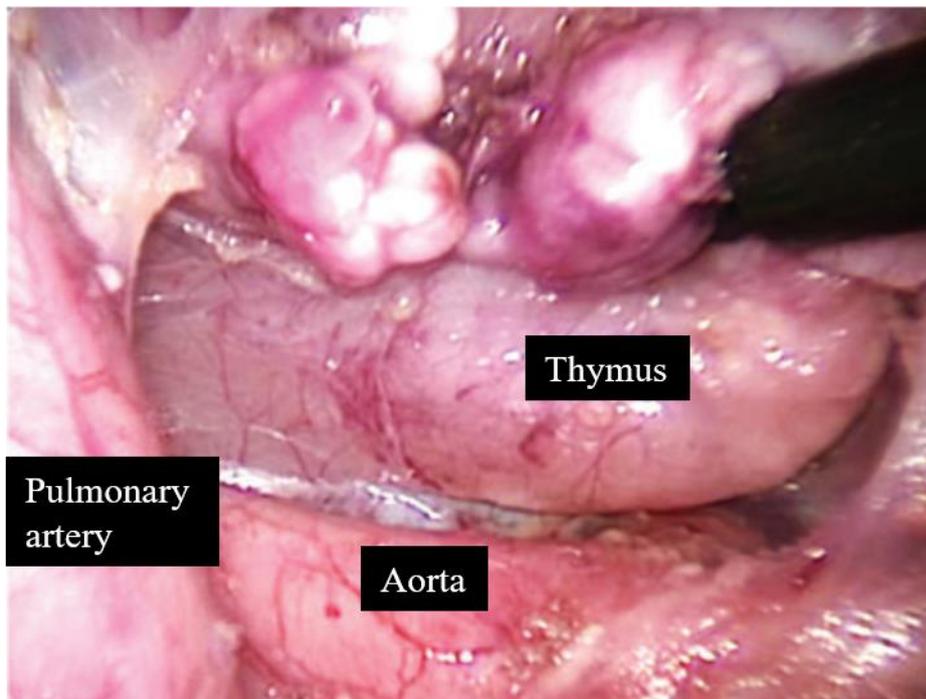


Figure 2.

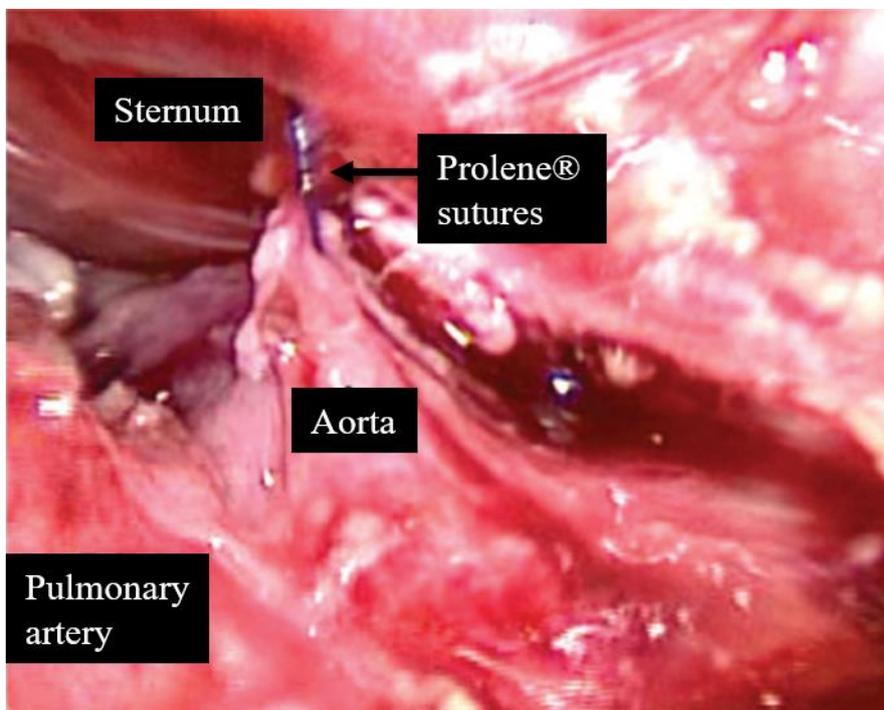


Figure 3.

