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Interactive Cardiovascular and Thoracic Surgery 33 (2021) 462–468
doi:10.1093/icvts/ivab100 Advance Access publication 8 May 2021

ORIGINAL ARTICLE

Cite this article as: Dolmaci OB, Fockens MM, Oomen MW, van Woensel JB, Hoekstra CEL, Koolbergen DR. A modified surgical technique for aortopexy in tracheobronchomalacia. Interact CardioVasc Thorac Surg 2021;33:462–8.

A modified surgical technique for aortopexy in tracheobronchomalacia

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Received 30 November 2020; received in revised form 11 February 2021; accepted 10 March 2021

Key question

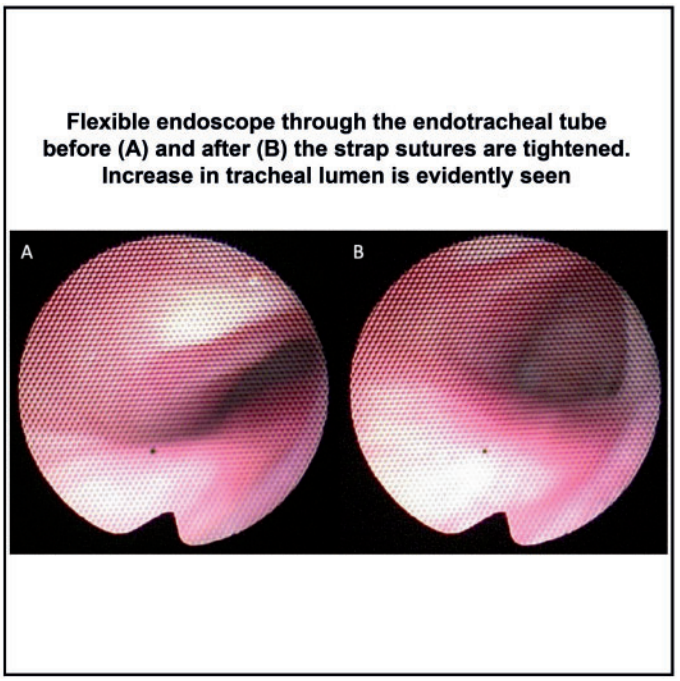
What is the outcome of a modified aortopexy with aortic arch strap sutures for treatment of tracheobronchomalacia?

Key finding(s)

Modified aortopexy was successful in 91.7%. Complications occurred in 8.3% and mortality was 4%.

Take-home message

Modified aortopexy with strap sutures is an effective and safe treatment for severe tracheobronchomalacia.



Abstract

OBJECTIVES: Tracheobronchomalacia (TBM) is characterized by collapse of trachea, bronchi or both, leading to dyspnoea, expiratory stridor, coughing or recurrent airway infections. Surgical treatment with aortopexy is warranted for severe TBM. We describe a modified aortopexy technique with aortic wall strap sutures that evenly distributes the traction force over the full width of the aortic arch. The aim of this study was to determine the outcomes of this modified anterior aortopexy technique.

METHODS: Retrospective chart review of all patients undergoing aortopexy with aortic wall strap sutures for TBM between January 2010 and June 2020 in 2 tertiary hospitals in the Netherlands.

[†]These authors shared first authorship and contributed equally.

RESULTS: Twenty-four patients [median age 9 months (interquartile range 2–117 months); 71% male] underwent aortopexy with the modified technique for TBM (52%), tracheomalacia (40%) or bonchomalacia (8%). Aortopexy was successful in 91.7%, defined as relief or decrease of respiratory symptoms and no need for respiratory support. Complications occurred in 8.3% and mortality was 4%.

CONCLUSIONS: Aortopexy with non-absorbable strap sutures seems an effective and safe treatment for severe TBM. This study supports the hypothesis that strap sutures provide a solid and reliable traction force, but future comparative studies should confirm the benefit of strap sutures over conventional techniques.

Keywords: Aortopexy • Tracheomalacia • Tracheobronchomalacia

ABBREVIATIONS

BM	Bonchomalacia
IQR	Interquartile range
PICU	Paediatric intensive care unit
TBM	Tracheobronchomalacia
TM	Tracheomalacia

INTRODUCTION

Tracheobronchomalacia (TBM) is a respiratory condition characterized by local or generalized collapse of the lumen of the trachea and main bronchi [1–3]. The trachea or main bronchi can also be affected independently [tracheomalacia (TM) or bronchomalacia (BM)] [4]. TBM can be classified as congenital (i.e. primary) or acquired (i.e. secondary) malacia [5]. In primary malacia, intrinsic collapse of the trachea occurs as a result of congenital weakness of tracheal cartilage [6]. It is often seen in children with oesophageal atresia with tracheoesophageal fistula. In secondary malacia, airway collapse regularly occurs as a result of extrinsic compression by vascular rings or slings [5, 7, 8]. Airway compression by the brachiocephalic artery (also known as innominate artery compression syndrome) is the most common cause of secondary malacia [9]. In literature, the distinction between primary and secondary malacia is not always made clear and often primary and secondary malacia are grouped together when discussing the topic. Furthermore, quite regularly a combination of both forms exist simultaneously [10].

Respiratory symptoms that may arise in neonates or children due to TBM include dyspnoea, wheeze, expiratory stridor, prolonged inspiratory or expiratory phase, abundant tracheal secretions, coughing, recurrent airway infections or pneumonias, and acute life-threatening events [10]. Rigid tracheobronchoscopy with the child breathing spontaneously is essential to confirm the diagnosis and to determine the location and extent of airway collapse. If secondary malacia is suspected upon tracheobronchoscopy, computed tomography-scanning of the thorax is performed to identify the cause of the compression [4, 11].

The treatment choice for TBM depends on the severity of symptoms. In patients with mild TBM, treatment may consist of saline nebulizers (for secretions thinning and airway clearance), low-dose corticosteroids inhalers (to decrease inflammation and secretions), ipratropium bromide inhalers (to decrease secretions) or antibiotics in case of recurrent airway infections [7]. In moderate TBM, supportive treatment may be needed such as oxygen supply, nasal high flow canula or nasal continuous positive airway pressure (CPAP) (to provide additional tracheal intraluminal pressure to distend the trachea during expiration and decrease expiratory resistance). In severe TBM, this supportive treatment may be insufficient to relieve respiratory symptoms and surgical intervention in the form of an aortopexy should be considered [4, 7]. Currently, the standard surgical treatment is an aortopexy combined with correction of concomitant vascular anomalies if necessary [12, 13]. Several surgical approaches have been described, including the anterior (with full or partial sternotomy), lateral and thoracoscopic approaches [13–15]. Traditionally, an anterior aortopexy is performed using single 'U' stitches on the aortic wall [16]. In this article, we propose a modified technique of aortopexy with aortic arch strap sutures. We assume that strap sutures distribute the traction force more evenly over the full width of the aortic arch, leading to improved airway patency and better surgical outcomes. The aim of this study was to determine the outcomes of this modified aortopexy technique.

PATIENTS AND METHODS

Study population

This retrospective cohort study was conducted at Amsterdam University Medical Center location AMC and Leiden University Medical Center, 2 tertiary hospitals in the Netherlands. All patients who underwent an aortopexy with the modified surgical method for secondary TM, BM or TBM as confirmed with rigid tracheobronchoscopy between January 2010 and June 2020 were identified from the surgical databases. All procedures were performed by one cardiothoracic surgeon (D.R.K.). Data on patient demographics, medical history, tracheobronchoscopy findings, surgical details and postoperative course were extracted from the electronic health records. Respiratory symptoms were scored pre- and postoperatively. Approval for this study was granted by the medical ethics committee and patient consent was waived.

Outcome measures

Primary outcome measure was successful treatment of respiratory symptoms by aortopexy, defined as complete or partial relief of respiratory symptoms and no need for respiratory support. Secondary outcome measures included relief of specific respiratory symptoms, length of paediatric intensive care unit (PICU) stay, complications including reoperations or other interventions, readmissions to the ward or PICU within 3 months after discharge and mortality.

Surgical technique

Aortopexy is performed through a median sternotomy. The thymus is (partially) resected in order to create more space in the anterior mediastinum. Subsequently, the pericardium is partially divided at the superior end to obtain a clear view of the ascending aorta and the aortic arch. Two non-absorbable PTFE (Bard®

PTFE braided tape, preferred in patients of >8 kg) or Mersilene™ (Ethicon ®, preferred in patient of <8 kg) straps are sutured to the adventitial layer of the aortic arch; one on the inner curvature and one on the outer curvature of the arch. The straps are attached on the aorta over a width of at least 1 cm per strap in order to evenly distribute the traction force over the full width of the aortic arch (see Fig. 1). The pericardium is generally approximated with 1 or 2 stitches. Then, the straps are attached just parasternal to either the left or right second rib, depending on which side optimally reduces tracheal compression without creating haemodynamic alterations. Typically, the sternum is closed with temporary or permanent sutures before pulling on the straps in order to evaluate the effect with normal geometry of the chest wall. The result of the aortopexy on the tracheal lumen is observed with a flexible endoscope through the endotracheal tube (see Fig. 2 and Video 1). Postoperatively, patients are observed at least one night at the intensive care unit. Extubation is preferably performed in the first 12–24 h after surgery.

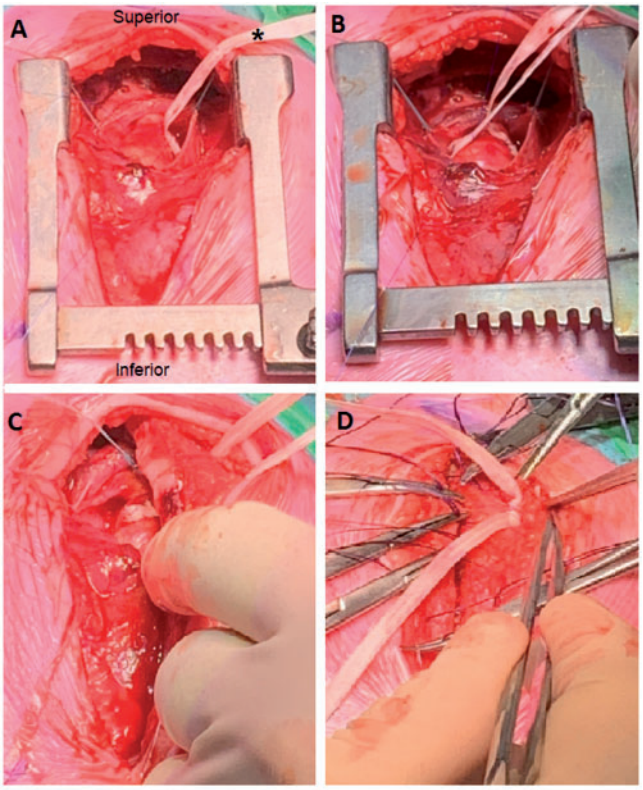


Figure 1: Aortopexy with non-absorbable strap sutures placed on the inner and outer curve of the aortic arch to evenly distribute traction force. (**A**) Placement of the left-sided PTFE strap (inner curve). (**B**) Placement of right-sided PTFE strap (outer curve). (**C**) Parasternal attachment of the PTFE straps. (**D**) Closure of the sternum. * = PTFE-strap (Bard® PTFE braided tape).

Statistical analysis

Continuous variables with a normal distribution are presented as mean ± standard deviation and non-normally distributed continuous variables as median and interquartile range (IQR). Categorical data are presented as frequencies and percentages. Skewness and kurtosis were examined for each variable. The Shapiro–Wilk test was performed to test the normality of the distribution of each variable. Two tests were used to analyse the effect of surgery on the pre- and postoperative symptoms, the McNemar's test for categorical variables and the Wilcoxon signed-rank test for continous (non-normally distributed) variables. A *P*-value of <0.05 was considered to be significant. All statistical analyses were conducted using IBM SPSS for Windows version 25.0.

RESULTS

Patient population

Baseline characteristics are displayed in Table 1. A total of 24 patients were included with a median postoperative follow-up of 25.5 months (IQR 18–34 months). The median age was 9 months (IQR 2–117 months) and 71% was male. Oesophageal atresia with a trachea-oesophageal fistula was the most common comorbidity (25%); all of these patients had undergone surgical correction prior to aortopexy. The innominate artery compression syndrome was the most common aetiology of TBM in 54.2% of the cases (Table 2). Prior to aortopexy, 6 patients (25%) were admitted to PICU with a median stay of 20 days (IQR 11–64 days). All

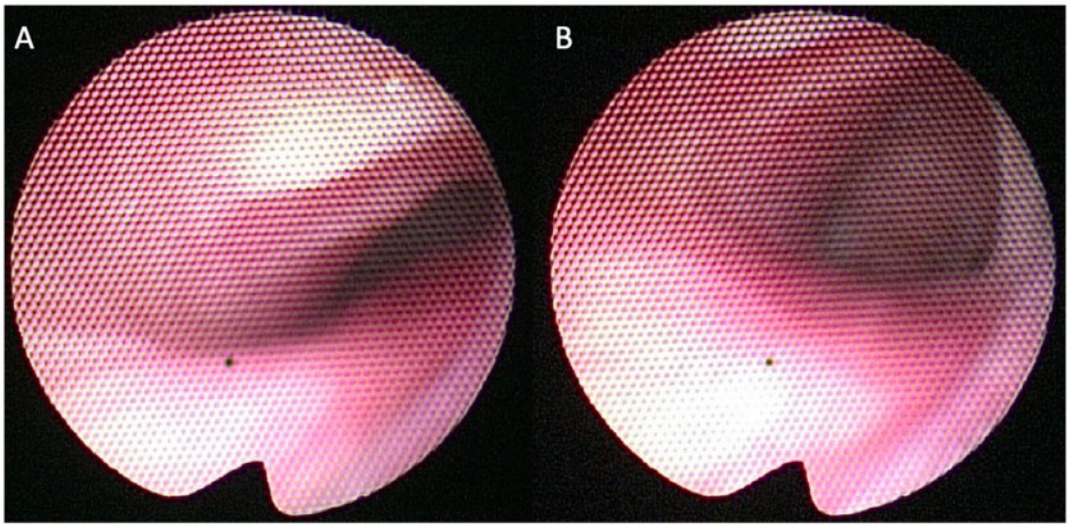
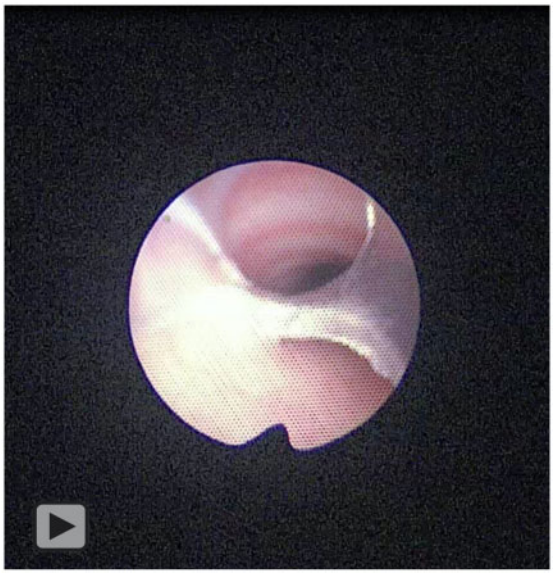


Figure 2: Flexible endoscope through the endotracheal tube before (**A**) and after (**B**) the strap sutures are tightened. Increase in tracheal lumen is evidently seen.



Video 1: Flexible endoscope through the endotracheal tube at the moment of strap sutures tightening in a patient with secondary tracheomalacia. Increase in tracheal lumen is evidently seen.

Table 1: Baseline characteristics

Characteristics	*n* = 24
Age (months)	9 (2–117)
Male	17 (70.8)
Weight (kg)	8.3 (4.57–26.78)
Premature	4 (16.7)
Gestational age of premature patients (weeks)	33 (26.25–33)
Comorbidities	
No comorbidities	15 (62.5)
Oesophageal atresia with tracheo-oesophageal fistula	6 (25)
VACTERL	2 (8.3)
Cystic fibrosis	1 (4.2)
Wilson's disease	1 (4.2)
Respiratory status prior to surgery	
Preoperative intubation	6 (25)
Tracheostomy	0
Medical treatment prior to surgery	
At least one course of AB treatment	13 (54.2)
Prophylactic AB treatment	9 (37.5)
Corticosteroid inhaler	5 (20.8)
Saline nebulizer	3 (12.5)
Salbutamol inhaler	3 (12.5)
Ipratropium inhaler	1 (4.2)
Airway malacia location	
Tracheobronchomalacia	13 (54.2)
Tracheomalacia	9 (37.5)
Bronchomalacia	2 (8.3)

Data are presented as *n* (%) or median (interquartile range).
AB: antibiotic.

of these patients were intubated because of respiratory insufficiency due to TBM.

Surgical characteristics

All patients underwent aortopexy through a median sternotomy as described above in the surgical technique section. Additional procedures were performed concomitant with aortopexy in 10 patients (41.7%): Kommerell's diverticulum resection in 9 patients (37.5%), resection of an atretic left aortic arch in 2 patients (8.3%), resection of an atretic right aortic arch in 1 patient (4.2%) and ventricular septal defect closure in 1 patient (4.2%). Cardiopulmonary bypass was used in 3 patients (12.5%) because of a planned concomitant procedure. Furthermore, a partial or subtotal thymectomy was performed in 20 patients (83.3%) and the ductus arteriosus was resected in 19 patients (79.2%).

Table 2: Vascular aetiology of secondary tracheobronchomalacia (*n* = 24)

Aetiology of tracheal stenosis	*n* (%)
Innominate artery compression syndrome	13 (54.2)
Right aortic arch, left ductal ligament	8 (33.3)
Double aortic arch	3 (12.5)
Right dominant	2 (8.3)
Left dominant	1 (4.2)

Clinical outcomes

Aortopexy with the modified technique was successful in treating respiratory symptoms in 22 patients (91.7%). Relief of respiratory symptoms was complete in 17 patients (70.8%) and partial in 5 patients (20.8%). Respiratory symptoms in 2 patients (8.3%) did not improve after aortopexy. Specific pre- and postoperative respiratory symptoms are displayed in Table 3. Dyspnoea and excessive cough were the 2 most common preoperative symptoms (58.3% and 45.8%, respectively). Statistical analyses showed a significant postoperative reduction of dyspnoea, wheezing, stridor, feeding problems, coughing, saturation dips and recurrent bronchitis. Furthermore, excessive salivary secretions, failure to thrive, tachypnoea, acute life-threatening events and recurrent pneumonia were observed in less patients after aortopexy, but this difference was not significant.

The median postoperative PICU length of stay was 1 day (IQR 1–4.75 days). Except for 3 patients, all patients were extubated within 24 h after surgery. Two of the 3 patients with a prolonged postoperative intubation time were already intubated preoperatively, which may explain a prolonged weaning period. The third patient suffered bronchospasms after aortopexy that required prolonged intubation. All 3 patients were successfully extubated within 5 days after surgery.

Complications, readmissions and mortality

Complications occurred in 2 patients (8.3%) due to the aortopexy. Both had a complication warranting intervention: one patient with a postpericardiotomy syndrome who developed pericardial effusion 4 weeks after surgery, which required pericardiocentesis, whereas the other patient developed a pneumothorax, which was treated with a pleural drain.

Following aortopexy, 2 patients (8.3%) were readmitted to the PICU: one patient was monitored after pericardiocentesis and another patient had dyspnoea and stridor after a choking incident for which brief treatment with nasal high flow canula therapy was given. One patient (4.2%) was readmitted to the paediatric ward after discharge because of a fever without an evident

Table 3: Specific pre- and postoperative respiratory symptoms ($n = 24$)

Characteristic	Preoperative	Postoperative	*P*-value
Dyspnoea	14 (58.3)	1 (4.2)	<0.001
Wheezing	8 (33.3)	1 (4.2)	0.016
Stridor	9 (37.5)	1 (4.2)	0.008
Feeding problems	9 (37.5)	3 (12.5)	0.031
Coughing	11 (45.8)	1 (4.2)	0.001
Tachypnoea	5 (20.8)	1 (4.2)	0.125
Saturation dips	7 (29.2)	1 (4.2)	0.031
Excessive secretions	7 (29.2)	3 (12.5)	0.125
Failure to thrive	2 (8.3)	1 (4.2)	1.000
ALTE	1 (4.2)	0	1.000
Pneumonia	5 (20.8)	2 (8.3)	0.375
Number of episodes*	7.29	5.88	0.141 (95% CI 0.239-0.256)
Bronchitis	7 (29.2)	1 (4.2)	0.031
Number of episodes*	8.20	7.84	0.059 (95% CI 0.122-0.135)

Data are presented as *n* (%) or per 100 patient-years (*).
ALTE: apparent life-threatening event.

infectious focus, which was treated successfully with broad-spectrum antibiotics.

Unfortunately, one patient died 1 month after surgery. This concerned a 3-month-old male neonate at the time of aortopexy. He was born prematurely at 32 weeks with a birth weight of 1330 g and had previously undergone surgical correction for oesophageal atresia with trachea-oesophageal fistula. Rigid tracheobronchoscopy showed severe TM with a tracheal lumen of ∼10%. Subsequently, the aortopexy was performed at the age of 2 months (weight 2725 g). During closure of the sternum, a decline in cardiac output was suspected as monitors showed diffuse ST-segment depressions and decreased carbon dioxide expiration. The actions of the anaesthesiologist (i.e. filling and inotropes) did not improve the decreased cardiac output. Cardiac output normalized after immediate revision of the aortopexy. Postoperative transthoracic ultrasound showed no abnormalities. Despite this, he developed epileptic seizures and an MRI scan performed 5 days after surgery showed extensive ischaemic brain damage. Because of a poor neurological prognosis and additional complications further treatment was considered futile and was withdrawn 1 month after aortopexy.

DISCUSSION

In recent years, aortopexy has proven to be the preferred treatment for children suffering severe TBM [13]. However, aortopexy is still unsuccessful in an estimated 20% of patients. Improvement of surgical technique should therefore always be pursued to improve the surgical outcome. This study shows that our modified anterior aortopexy technique is an effective and safe treatment for severe TBM.

Several surgical approaches of aortopexy have been described for the management of TBM [13]. Anterior aortopexy is traditionally performed using 2 double-needle 'U' stitches, which are placed on the origin of the innominate artery [16]. This technique was previously used in our institute as well. Our study shows the result of a modified technique of the anterior aortopexy, which is performed using 2 non-absorbable straps instead of 2 single stitches. The hypothesis behind this modified technique is that using 2 straps leads to an even distribution of the traction force over the full width of the aortic arch. Dividing the traction force over the full width of the aortic arch, in turn, leads to a more effective and more reliable traction force. Interestingly, the effect of the strap sutures can be visualized on postoperative imaging as these sutures are non-absorbable (Fig. 3).

Aortopexy is considered a relatively safe and effective treatment for TBM. A review of 40 studies on aortopexy for TBM showed that 82% of patients experienced satisfactory improvement, ranging from 41.7% in the study with the least effect to 100% in studies with the best effect [13]. In our study, 91.7% of patients with TBM showed relief or improvement of respiratory complaints as a result of aortopexy with the modified technique. This result is higher than 80% improvement reported in the aforementioned review [13]. In this review, only 6 other studies with at least 10 patients showed more than 90% improvement. As this review did not separately report complete and partial effect, it is not possible to further compare our results of complete relief (70.8%) and partial effect (20.8%). Furthermore, in literature a lack of effect or worsening of complaints is described in ∼12% [13]. We report a lack of effect in 8.3%. In our patients, this is attributed to cystic fibrosis in one patient and the decease of another patient as described earlier.

The overall incidence of postoperative complications, as described in the literature, is 16.6% [13]. The most common complications are related to the lungs (e.g. pneumothorax or atelectasis) and postoperative pleural or pericardial effusion. The current study reported complications in 8.3% of patients, which is in accordance with literature. Periprocedural mortality of aortopexy varies between 0% and 16% [13, 17, 18]. In our study cohort, one patient (4%) died as a result of the withdrawal of therapy because of postoperative brain damage with a poor prognosis. Unfortunately, we were unable to identify the exact cause of ischaemic brain damage in this patient. Furthermore, we did not identify any articles reporting ischaemic brain damage following aortopexy. It remains therefore unknown whether the decease of our patients was related to our modified aortopexy technique.

This study is limited by a relatively short follow-up time, the retrospective aspect, single-centre nature, and the lack of a comparative cohort of patients. Although the follow-up period of this study was insufficient to draw conclusion about the long-term outcomes, we have no reason to assume that our modified

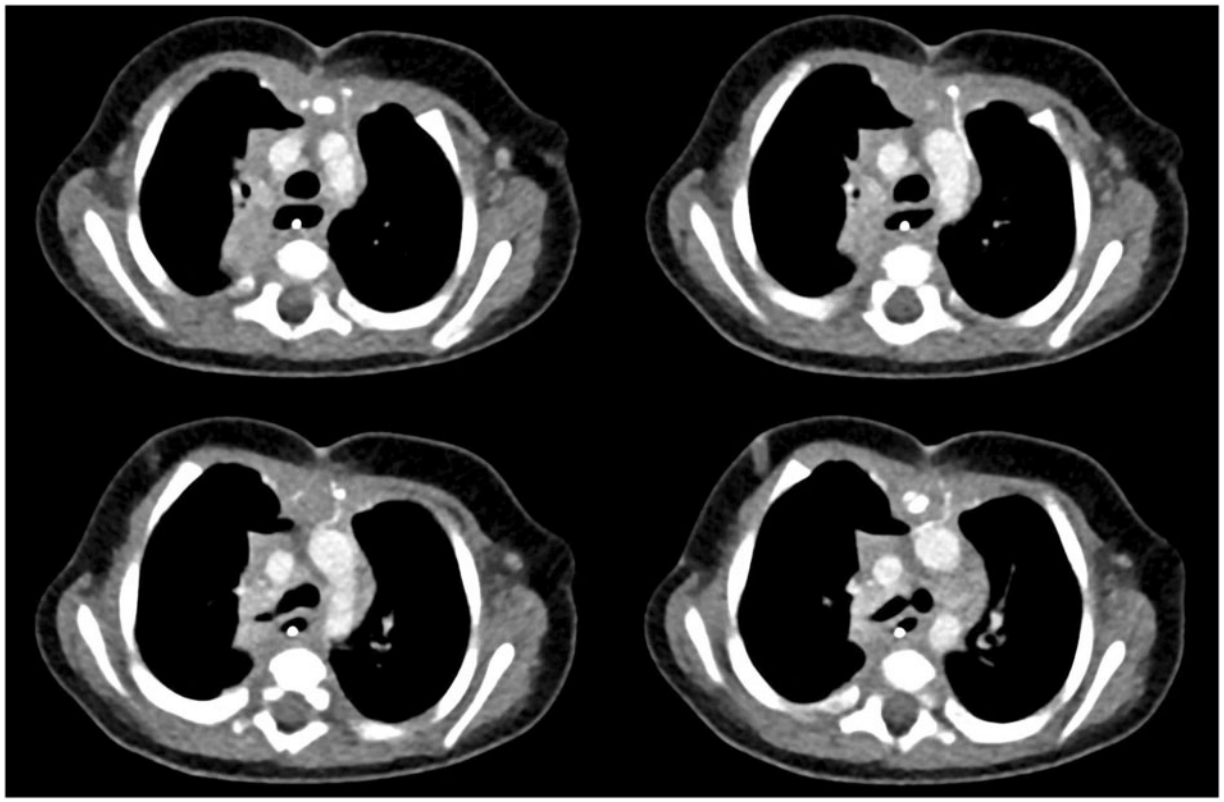


Figure 3: Computed tomography scan of the thorax after aortopexy showing the PTFE straps. The non-absorbable straps run from the aortic arch to the left parasternal attachment.

technique with strap sutures would be inferior in the long term to the traditional single stitches. Future prospective studies are required to confirm the results of this study.

CONCLUSION

Aortopexy with non-absorbable strap sutures is an effective and safe treatment for severe TBM, as it was successful in 91.7% and led to complications in only 8.3%. This study endorses the hypothesis that strap sutures provide a solid and reliable traction force, but future comparative studies should confirm the benefit of strap sutures over conventional techniques.

Conflict of interest: none declared.

Author contributions

Onur B. Dolmaci: Data curation; Investigation; Methodology; Writing—original draft. **M. Matthijs Fockens:** Data curation; Investigation; Methodology; Writing—original draft. **Matthijs W. Oomen:** Writing—review & editing. **Job B. van Woensel:** Writing—review & editing. **Carlijn E.L. Hoekstra:** Methodology; Supervision; Writing—review & editing. **Dave R. Koolbergen:** Conceptualization; Supervision; Visualization; Writing—review & editing; Invention of surgical technique.

Reviewer information

Interactive CardioVascular and Thoracic Surgery thanks Christoph Schmitz, Kerem M. Vural and the other anonymous reviewer(s) for their contribution to the peer review process of this article.

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