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Thoracoscopic Aortopexy for Tracheomalacia

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Abstract

Background Tracheomalacia is a condition that may result in acute life-threatening events. Aortopexy has become a generally accepted mode of treatment, although the impact of a thoracotomy is considerable. With the advent of minimal invasive surgery the impact of such a procedure may be greatly reduced. This study evaluates the outcome of thoracoscopic aortopexy for tracheomalacia.

Methods Retrospective analysis of a database containing all patients with thoracoscopic aortopexy. Based on symptomatology, on indication 24 h pH study and/or rigid tracheo-bronchoscopy were carried out to confirm the diagnosis. A three-trocar technique was used on the left side together with intra-operative flexible tracheoscopy through the tube. Follow-up continued for the duration of the study.

Results Between January 2002 and December 2012, 16 patients presented with severe tracheomalacia. Fourteen of them had a history of esophageal atresia. Age at time of aortopexy varied from 2 weeks to 12 months. All procedures were performed thoracoscopically without intra-operative complications. In five children, there was a recurrence of symptoms within 2–4 weeks for which a re-thoracoscopic aortopexy was carried out successfully. With a follow-up of 6 months to 10 years all patients are thriving and are without incidents.

Conclusion This is the largest single-center series of thoracoscopic aortopexy for tracheomalacia so far. All procedures were completed successfully without complications. Although recurrences occur, as in open surgery, the ultimate results are satisfactory. In an era of minimal invasive surgery the thoracoscopic approach is feasible and safe, even in redo-procedures.

Introduction

Tracheomalacia is a rare condition that is most frequently encountered in children with esophageal atresia, although there is a spectrum varying from mild symptoms like the well-known seal cough to acute life-threatening events (ALTE) with sometimes fatal outcome. Aortopexy has become the established treatment for dealing with severe tracheomalacia [1]. As a thoracotomy is still a major procedure, in the past an antireflux procedure was often performed first to exclude aspiration as a cause for the ALTE [1, 2].

With the advent of minimal invasive surgery, the impact of such a procedure may be greatly reduced.

So far only a limited number of anecdotic series of thoracoscopic aortopexy have been published [3–8]. In this

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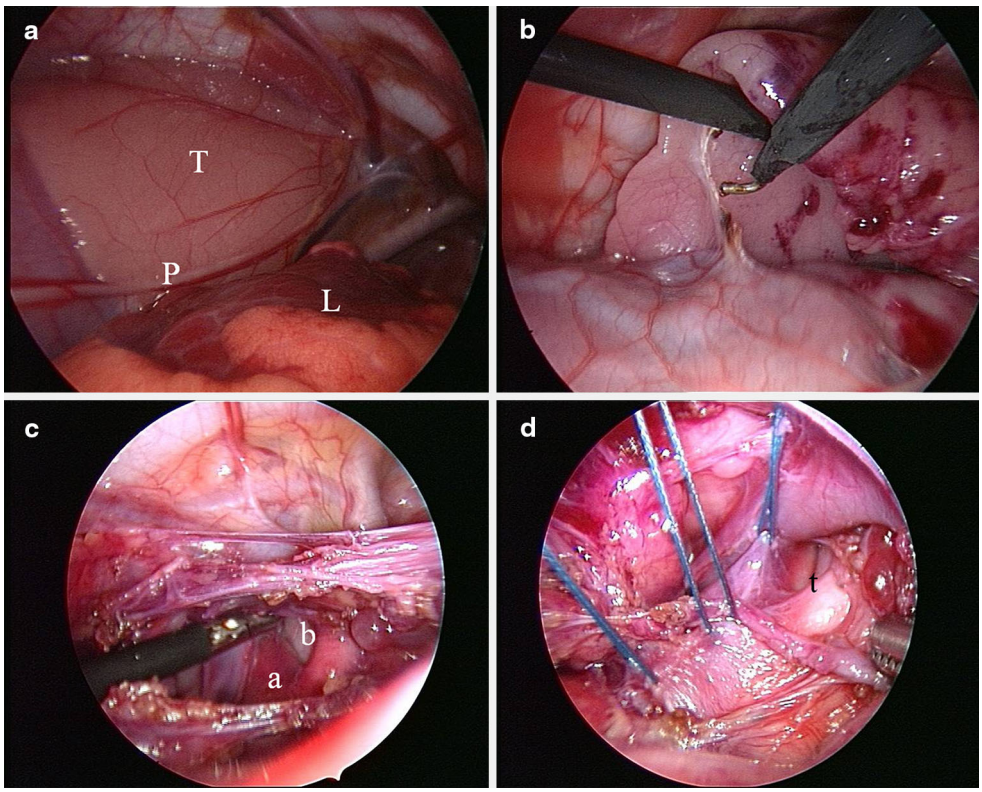

Fig. 1 **a** Overview of left thorax. *T* thymus, *P* phrenic nerve, *L* lung. **b** Mobilization of thymus. **c** Preparation of aorta (*a*) and bracheocephalic artery (*b*). **d** Overview after placement of traction sutures. Under control of a flexible scope (*t*) the effect of elevation is tested

article, we describe 10 years of experience with thoracoscopic aortopexy.

Materials and methods

Between January 2002 and December 2012, 16 patients presented with severe symptoms of tracheomalacia. Symptoms consisted of life-threatening events, recurrent infections or cyanotic spells, and apnea.

In case the cause was not clear the diagnosis was confirmed by 24 h pH study and rigid tracheo-bronchoscopy under spontaneous breathing. In case of massive gastro-esophageal reflux a laparoscopic antireflux procedure was undertaken first. However, if during trachea-bronchoscopy the anterior and posterior wall touched, there was an indication for primary thoracoscopic aortopexy.

There were 8 boys and 8 girls. Age varied from 2 weeks to 12 months ($M = 5$ months). Weight was between 3 and 9 kg (M-5.5 kg). Fourteen children had a history of esophageal atresia.

Surgical procedure

The technique was described earlier in the report of our first six cases [5], but has been adjusted since. Therefore, a description with the adjustments follows below. The child is placed in a supine position at the left side of the table with a tilt of the left shoulder to facilitate the approach of the anterior mediastinum from the left side. One 6 mm trocar for the camera and two 3.5 mm trocars for instrumentation are used. The left hemithorax is insufflated with CO2 with 5 mmHg and a flow of 2 l/min (Fig. 1a). Under identification of the phrenic nerve the pleura is opened, the left thymus lobe is mobilized (Fig. 1b), and the thymus is pushed to the right side of the mediastinum. Sometimes, it may be necessary to partly mobilize the right thymus in order to create sufficient room for the left lobe to be pushed over to the right side. The pericardium, the ascending aorta, and brachiocephalic artery can then be identified and prepared where necessary to give adequate access for placing the sutures (Fig. 1c). By pushing on the sternum and passing a 24G needle between the ribs next to the sternum the best location for the sutures can be determined. Small stab wounds are made and 3 × 0 Ethibond® (Cincinnati, OH) nonabsorbable sutures can be introduced through the sternum. We nowadays use a Reverdin's needle (Fig. 2a), because it is strong and the needle to hitch the aorta remains sharp. After hitching up the aorta adventitia the suture can be withdrawn with the use of an Endoclose® (TYCO Medical, Mansfield, MA) (Fig. 2b) introduced through the same stab wound but guided subcutaneously directly parasternally. Three to four trans-sternal sutures can be placed from the pericardium up to the

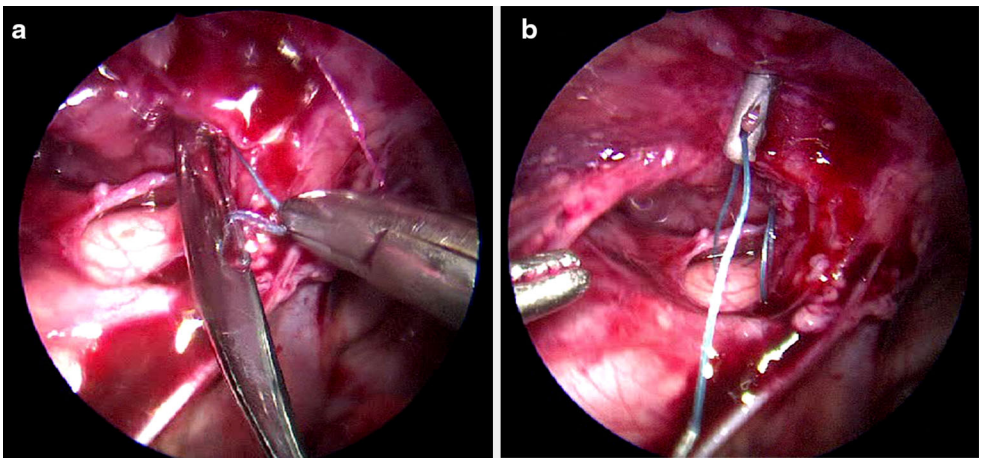

Fig. 2 **a** Introduction through the sternum of a Reverdin needle with a 3 × 0 Ethibond® suture in the patient with the dissection of the adventitia of the aorta, requiring a redo-aortopexy. **b** After hitching up the aorta wall with the use of an Endoclose® introduced parasternally, the suture is retrieved and pulled out for tying subcutaneously

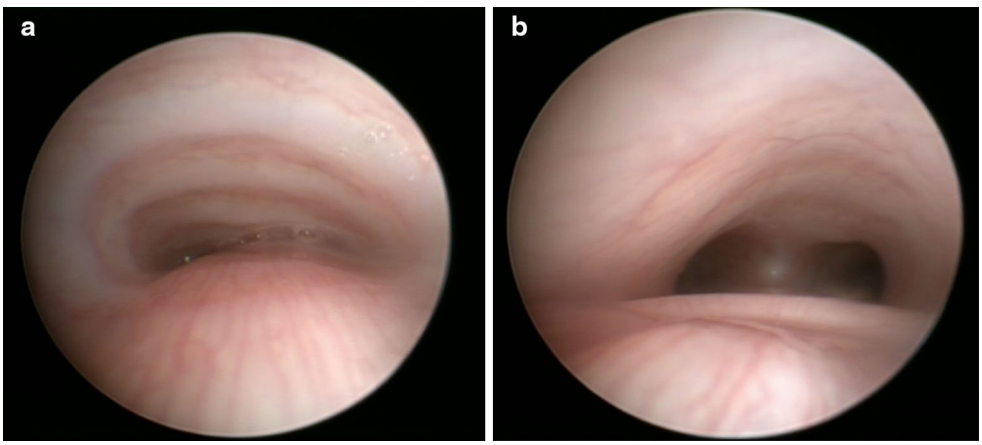

Fig. 3 **a** View of trachea before lifting up the aorta. Ventral and posterior wall are touching. **b** View of trachea after aortopexy

brachiocephalic artery (Fig. 1d). After the sutures are placed, flexible trachea-bronchoscopy through the tube is performed to observe the effect of pulling the aorta up against the posterior surface of the sternum (Fig. 3a, b) where necessary additional sutures are placed. The sutures are tied subcutaneously (Fig. 4a). At the end of the procedure, the pneumothorax is evacuated under direct vision. The trocar holes and stab wounds are closed with either absorbable Vicryl 4–0 or 5–0 sutures and Steristrips (Fig. 4b). No drain is left in place. Principally, extubation on the table is initiated unless there was extensive manipulation of the trachea during tracheoscopy. If the anesthesiologist deems it necessary, the children are kept in the intensive care unit overnight.

Central hospital permission was granted for performing this retrospective study.

Results

A laparoscopic antireflux procedure was initially performed in the two first patients because of suspected reflux. As symptoms did not subside, a thoracoscopic aortopexy was subsequently carried through successfully. All other patients demonstrated clear tracheomalacia during tracheoscopy with the posterior and anterior wall touching (Fig. 3a), and a primary thoracoscopic aortopexy was carried through.

All patients tolerated the thoracoscopic aortopexy well and no intra-operative complications were encountered.

At the end of the procedure with patient no. 16 (Table 1), there was some doubt as to the effect of the thoracoscopic aortopexy at the cranial end due to the tube that the anesthesiologist would not withdraw any further

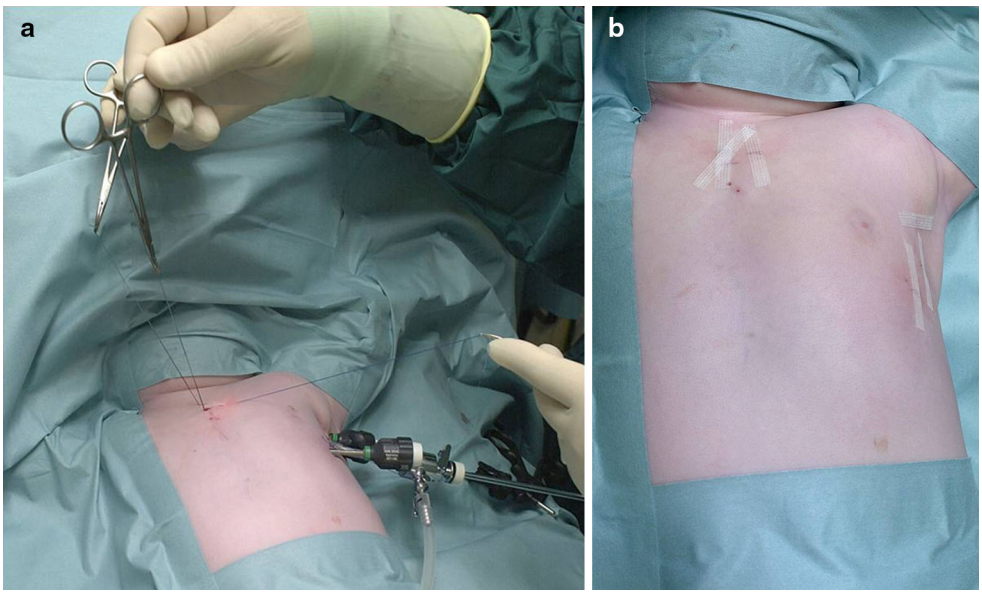

Fig. 4 **a** Under direct vision of both the endoscope and flexible tracheoscope the sutures can be lifted to view the effect of aortopexy. If the result is adequate the sutures are tied subcutaneously. **b** End result after closing stab wounds

for determination of the effect of the aortopexy. The child could not be extubated and at rigid tracheoscopy 2 days later it was evident that part of the trachea above the level of the manubrium still collapsed. Therefore, a suprasternal incision was made and additional sutures, directly placed at the anterior tracheal wall, lifted the residual ventral wall sufficiently. The child could be extubated the next day. One other child (no. 13) was born with esophageal atresia and aplasia of the right lung. The child developed severe tracheomalacia with hypoxic episodes. Even in this child it was possible to perform a thoracoscopic aortopexy under mild insufflation of 0.5 l/min and a pressure of 3 mm Hg. There was an immediate recovery postoperatively. None of the patients needed to stay on the ventilator. There were no more incidents of apnea or hypoxic spells. In most of the children there was no audible breathing anymore. All patients were discharged from the hospital between 2 days and 8 weeks. Indications for prolonged hospitalization were concomitant anomalies requiring additional treatment (Table 1). During follow-up, five children displayed recurrence of symptoms 2–4 weeks after the primary procedure requiring a redo-thoracoscopic aortopexy. It was remarkable how little adhesion formation had occurred and it was easy to place new or additional sutures where necessary. In one patient (no. 14) there was dissection of the adventitia from the aortic wall. Sutures had to be placed in the aortic wall itself. Fortunately this did not have adverse side effects. An overview of the causes for relapse is displayed in Table 2. Apart from the five patients that suffered from a relapse of symptoms due to recurrences, there have been no re-admissions for tracheomalacia-related complications. Parents indicated that they received a new child.

Follow-up is between 6 months and 10 years. All children are without further incidents (Table 1).

Discussion

When open surgery was performed the aortopexy was considered a major operation and usually, if the diagnosis was not totally clear, an antireflux procedure was performed first [1, 2]. In our series the first two patients also initially underwent a laparoscopic antireflux procedure, because we were not fully comfortable with the thoracoscopic approach. Subsequently only one other patient had an antireflux procedure because of ALTES due to 40 % reflux, and did not require additional aortopexy afterward. It is, therefore, always good to perform both a 24 h pH study and a rigid trachea-bronchoscopy during the diagnostic work-up if the cause is not clear. The four children who underwent a laparoscopic antireflux procedure later on did not have life-threatening events, but reflux-related problems like esophagitis or stenosis. A rigid bronchoscopy is mandatory, contrary to a flexible bronchoscopy, to better determine the exact extent of the malacia and to (again) exclude a proximal esophagotracheal fistula. In spite of explicit assurance by the referring physician that there was no proximal fistula on flexible endoscopy on two occasions, an additional proximal fistula was diagnosed and dealt with (Table 1).

Table 1 Patient characteristics

Patient no.	Age	Underlying disease	Symptoms	Duration operation	Intra-op. complications	Hospitalization	Postop. complications	Redo-procedure	Follow-up	Remarks
1	7 months	Esophageal atresia	ALTE	120	No	4 days	No	No	10 years, ARP at 5 months Good	
2	12 months	Esophageal atresia	ALTE	120	No	2 days	No	No	10 years, ARP at 6 months. Good	
3	3.5 months	Esophageal atresia	ALTE	100	No	12 days	ALTE	4 weeks	9 years, ARP at 12 months. Recurrent LAI	
4	4.5 months	Esophageal atresia	ALTE	150	No	40 days	RS pneumonia	No	9 years, ARP at 12 months. Good	
5	2 weeks	Esophageal atresia	ALTE	90	No	2 days	No	2 months	8 years Good	
6	4 months	Esophageal atresia	ALTE	200	No	13 days	Convulsion	No	7 years, ARP at 11 months. Good	
7	10 months	Esophageal atresia	Cricoid stenosis, broncho-tracheomalacie	120	No	6 days	No	No	6 years, feeding difficulties. ARP at 4 years	
8	11 months	Unknown	ALTE	130	No	16 days	ALTE	2 weeks	4 years. Good	
9	7 months	Unknown	ALTE	120	No	1 days	No	No	3 years. Good	
10	3 months	Esophageal atresia	ALTE	150	No	8 days	No	No	2 years. Good	ARM
11	4 months	Esophageal atresia	ALTE	120	No	8 days	Fausse route gastrostomy	No	2 years, lost to follow-up abroad	CHARGE
12	6 months	Esophageal atresia	ALTE	120	No	12 days	No	No	1, 5 years. Good	
13	8 months	Esophageal atresia	ALTE	90	No	3 days	No	No	1 year. Good	Single lung
14	4 weeks	Esophageal atresia	ALTE	90	No	6 weeks	ALTE	2 weeks	1 year. Good	ARM
15	2, 5 months	Esophageal atresia	ALTE	60	No	8 weeks	ALTE	5 weeks	1 year Good	Long gap EA, feeding difficulties
16	5 months	Esophageal atresia	ALTE	100	No	12 days	Not high enough		6 months. Good	Suprasternal additional sutures

ALTE acute life-threatening events, *RS pneumonia* respiratory syncytial pneumonia, *ARP* antireflux procedure, *ARM* anorectal malformation, *CHARGE* coloboma of the eye, heart defects, atresia of the nasal choanae, retardation, genital abnormalities, and ear abnormalities and deafness; *Long gap EA* long gap esophageal atresia, *LAI* lower airway infections

Table 2 Causes for recurrence

No. patient	Age of recurrence	Cause of recurrence
3	4w	Two additional sutures higher up at level bracheocephalic artery
5	2m	Dehiscence with disappearance of sutures. New sutures
8	2w	Tearing of sutures. Also additional sutures higher up
14	2w	Dehiscence of adventitia. New sutures in aortic wall.
15	5w	Dehiscence of sutures. New sutures placed.

De Cou et al. [8] were the first to describe the thoracoscopic aortopexy in 2001. Since then only seven publications have described the thoracoscopic approach for tracheomalacia [3–9]. So far our study is the largest series of thoracoscopic aortopexy reported from a single center. The major advantage is avoiding a large thoracotomy with all its sequelae, i.e., wound pain, scar formation, developmental, and postural anomalies. Good collaboration with the anesthesiologist is necessary. There is no indication for selective intubation and with moderate insufflation of max. 5 mm Hg and a flow of 2 l/min there is a good desufflation of the left lung. There are centers that approach the aortic arch from the right thorax, because they say the right thymus is smaller [4]. We have had no problems moving the thymus to the right hemithorax, with sometimes some mobilization of the right lobe of the thymus as well to push the left lobe out of the way sufficiently. We have had no indications for resecting part of the thymus. Good collaboration with the anesthesiologist is important, not only to monitor oxygenation, but also to give a total overview of the trachea during tracheoscopy. In the last patient, the anesthesiologist did not want to withdraw the tube far enough for a good overview of the upper part of the trachea and residual tracheomalacia in the upper part of the trachea was missed. This was approached from the neck 2 days later. In the total series, this case was noted as an insufficient procedure.

Recovery is remarkable. Older children can usually be extubated in the OR and transferred to the recovery room. Most of the children (15/16) have no audible breathing any longer. On one occasion, the parents asked if the child could be taken home from recovery because she was doing so well. It was deemed sensible to keep her overnight. Neonates are transferred to be extubated in the ICU in the evening or next morning, particularly if there was extensive manipulation of the trachea.

The fact that recurrences occur is probably related to the amount of tissue taken into a bite of the adventitia of the aorta. There is a subtle balance between not taking too deep a bite resulting in bleeding or too superficial a bite that will cause tearing. The case where the adventitia was dissected from the aortic is exemplary. Major concern is bleeding from the stitches. Conversion to open surgery may not be quick enough to save the child in case of major bleeding. However, such a calamity would only occur if the bites have been taken very deep and the suture is torn out. If some minor bleeding occurs, it suffices to press on the spot with an endoscopic instrument for a minute to let clotting take place. The suture can usually be left in place and used for the aortopexy. It can be discussed to use pledgets in the sutures. We have not done this to date, but in case of dissection of the adventitia it could be well indicated. In open surgery, relapse is described to occur in 5–20 % [10–12]. The recurrence rate in this series (37, 5 %) probably also reflects a learning curve. Jennings et al. describe 38 % recurrence [9] in their thoracoscopic series of seven patients. The thoracoscopic recurrence rate so far is higher than in open surgery, but we believe that we have now gone through our learning curve and have solid agreements with our anesthesiologists.

Conclusion

This is the largest single-center series of thoracoscopic aortopexy for tracheomalacia so far. All procedures were completed successfully without complications. Although recurrences do occur, as in open surgery, the ultimate results are encouraging to proceed with this procedure. In an era of minimal invasive surgery the thoracoscopic approach is feasible and safe, even in redo-procedures. Due to the limited number of cases, it is advisable to centralize the care of these children.

Conflict of interest David C. van der Zee and Marieke Straver have no conflict of interest.

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