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A case series perspective

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Published in:
Acta Paediatrica, International Journal of Paediatrics

DOI:
10.1111/apa.17452

Publication date:
2025

Document version:
Final published version

Received: 20 June 2024 | Revised: 22 September 2024 | Accepted: 26 September 2024

DOI: 10.1111/apa.17452

ACTA PÆDIATRICA NURTURING THE CHILD WILEY

CLINICAL OVERVIEW

Tracheomalacia and surgical options: A case series perspective

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Abstract

Tracheomalacia causes considerable morbidity in children, and the best treatment options remain debated. This paper presents a case series of seven Danish children who underwent surgical interventions, such as tracheopexy and aortopexy, demonstrating favourable clinical outcomes, notably with early intervention. We discuss the indications, timing and potential benefits of surgery for tracheomalacia in reducing respiratory symptoms caused by tracheal collapse. Our case series highlights the potential of surgical options in managing tracheomalacia, emphasising the need for standardised protocols, multidisciplinary and international collaboration, and further research to optimise treatment strategies and outcomes.

KEYWORDS
aortopexy, oesophageal atresia, surgery, tracheomalacia, tracheopexy

Congenital tracheomalacia is characterised by excessive collapsibility of the trachea, primarily due to a malformation of its horseshoe-shaped cartilage rings. This abnormality disrupts the tracheal structure, resulting in a flattened, wide anterior wall and a flaccid posterior wall that intrudes into the tracheal lumen, causing dynamic collapse during breathing. Additionally, the malformed cartilage makes the trachea prone to compression from nearby thoracic structures, such as the aortic arch.[1]

Tracheomalacia symptoms vary in severity, from milder manifestations such as wheezing, stridor and coughing with recurrent or severe lung infections[2] to more concerning episodes. These include brief resolved unexplained events (BRUEs) marked by changes in breathing, colour, muscle tone, or responsiveness or severe apparent life-threatening events with apnoea or respiratory arrest.[1,3,4]

Despite tracheomalacia occurring in at least 1 in 2100 children,[5] and being capable of causing life-threatening events, treatment

Abbreviations: AP, aortopexy; BRUE, brief resolved unexplained event; EA, oesophageal atresia; TP, tracheopexy.

strategies remain debated. Surgical intervention, notably tracheopexy (TP), has gained increasing international interest in recent years.[6] TP addresses the posterior wall intrusion by anchoring it to the anterior spinal ligament with a few non-absorbable sutures to prevent collapse.[7] Initially, TP was developed to prevent recurrence of tracheoesophageal fistula following oesophageal atresia (EA) repair. Given the high association of tracheomalacia with EA, seen in up to 86.7% of cases,[8] TP showed a significant positive impact on respiratory issues.[9] Aortopexy (AP) is another surgical option, involving forward pulling and securing parts of the aorta to the sternum with sutures, typically requiring removing the thymus to create space.[1,10] External splinting may be considered for highly selected, complex cases to support the tracheal walls, although there are concerns about erosion into surrounding structures and infection.[6]

Rigshospitalet in Copenhagen, Denmark, collaborated with Great Ormond Street Hospital in London, UK, to offer surgical intervention for severe tracheomalacia in Danish paediatric patients. Since the onset of the COVID-19 pandemic, Rigshospitalet has performed several surgical procedures independently.

In Denmark, severe respiratory symptoms from tracheomalacia and the need for surgical interventions have highlighted the challenges faced by children with tracheomalacia and EA. Recognising the lack of Nordic literature on tracheomalacia management, we initiated a large Danish study to provide a comprehensive assessment. This paper focuses on a subgroup from the national study: patients who underwent surgical intervention for tracheomalacia. By reviewing seven cases, we discuss the indications, optimal timing and clinical outcomes of surgical interventions for tracheomalacia, aligning with the objectives of the ongoing Danish study.

We conducted a retrospective clinical review of seven children who underwent surgery for tracheomalacia at hospitals in Denmark and London, between August 2019 and September 2022. Data were extracted from medical records, with informed consent.

The general demographics and clinical data of the patients are summarised in Table 1. Below, we present individual case details.

Patient 1 was a female infant born full term who underwent EA repair at 2 days old. She experienced three episodes of respiratory arrest, the first occurring at 6 weeks of age, along with multiple instances of respiratory insufficiency, often triggered by meals. Bronchoscopy confirmed severe tracheomalacia with kissing walls in the distal trachea. Initial AP surgery, performed in Denmark when she was 7.5 months old, failed to alleviate respiratory symptoms, and she experienced another respiratory arrest 3 weeks later. Subsequent bronchoscopy revealed persistent severe tracheomalacia. Five months after AP surgery, she suffered a cardiac arrest but quickly regained spontaneous circulation while intubated. At 12 months old, she underwent TP and a second AP with a Gore-Tex splint in the distal trachea at Great Ormond Street Hospital. Significant improvement in breathing was noted 5 days postoperatively, with increased energy during play. However, 1 month later, she experienced two episodes of respiratory insufficiency, one requiring admission and intubation. Bronchoscopy revealed some persistent intrusion of the posterior wall and a distal elliptical defect, but overall tracheal lumen was improved. She was extubated the same night and discharged. No further respiratory arrests occurred during the 4.6-year follow-up.

Key notes

- Congenital tracheomalacia involves excessive trachea collapsibility due to malformed cartilage, leading to breathing issues and vulnerability to thoracic structure compression.
- Symptoms range from mild wheezing to severe life-threatening events, and while prevalent in 1 in 2100 children, treatment strategies, especially surgical, remain debated.
- Surgical options like tracheopexy and aortopexy are explored, with variable success rates and ongoing discussions about optimal intervention timing and methods.

Patient 2 was a male infant born at term. EA was diagnosed and repaired on his first day of life. At 6 months, he experienced episodes of respiratory collapse related to viral pneumonia, complicated by pre-existing tracheomalacia, which required 11 days of extracorporeal membrane oxygenation. At 7 months, he underwent TP surgery at Rigshospitalet, which included resection of a tracheal pouch and a newly identified proximal tracheoesophageal fistula. He remained intubated for 40 days. Due to recurrent apnoea, an additional AP was performed 3 weeks after the TP, resulting in significant improvement in tracheal patency. Postoperative complications included atelectasis. The patient received continuous positive airway pressure treatment at home twice daily for 9 months. He remained free of respiratory issues throughout the 4-year follow-up period.

Patient 3 was a male infant born to term who underwent EA repair on his first day of life. He subsequently experienced apnoea and respiratory arrests, leading to bronchoscopy at 2 months, which revealed severe tracheomalacia with over 90% collapse in the distal trachea. After reviewing the bronchoscopy and CT scan, Great Ormond Street Hospital recommended combined AP and TP. However, due to COVID-19 restrictions, transfer was not possible. As respiratory arrests and BRUEs escalated, AP surgery was performed at Rigshospitalet at 4 months old. Although the AP procedure significantly reduced respiratory symptoms, severe tracheomalacia persisted with near-collapse of the posterior wall and tracheal stenosis. Consequently, TP was performed 22 days later, resulting in substantial improvement of the tracheal lumen. Surgical complications included subcutaneous emphysema, but no postoperative respiratory support was needed. During the 3.8-year follow-up, no respiratory issues or hospital admissions were reported.

Patient 4 was a 6-year-old female, born full term, diagnosed with tracheomalacia and tracheal stenosis at 18 months. Bronchoscopy showed a flattened anterior wall and an intruding posterior wall affecting tracheal segments. She experienced dyspnoea, rapid exhaustion, and recurrent respiratory infections, leading to multiple hospital

TABLE 1 General demographics and clinical data.

Patient number	Basic disease	Sex (M/F)	Preoperative TM symptoms	Age 1st intervention	First surgical procedure	Age additional intervention	Additional procedure	Complications	Follow-up period (years)
1	EA[a]	F	Respiratory arrest, respiratory insufficiency, cardiac arrest	7.5 months˙	AP[d]	12 months˙	AP[d] + TP[e] + external splint	-	4.6
2	EA[a]	M	Respiratory collapse, severe respiratory infections, apnoea	7 months˙	TP[e]	8 months˙	AP[d]	Atelectasis	4
3	EA[a]	M	BRUE[c], respiratory arrest, apnoea	4 months˙	AP[d]	5 months˙	TP[e]	Subcutaneous emphysema	3.8
4	Tracheal stenosis	F	Physical limitation, recurrent respiratory infections, dyspnoea	6 years	AP[d] + TP[e]	-	-	-	3.5
5	CF[b]	M	BRUE[c], apnoea, noisy breathing	5 months	AP[d] + TP[e] + external splint	-	-	-	2.7
6	EA[a]	F	BRUE[c]	3 months˙	AP[d]	4 months, 4.5 months˙	AP[d], TP[e]	CVC[f] infection	1.9
7	EA[a]	M	Physical limitation, respiratory difficulties eating	14 years	TP[e]	-	-	Pneumothorax, atelectasis	1.6

[a]Oesophageal atresia.
[b]Cystic fibrosis.
[c]Brief resolved unexplained event ˙Months.
[d]Aortopexy.
[e]Tracheopexy.
[f]Central Venous Catheter.

admissions. At 6 years old, she underwent simultaneous TP and AP at Great Ormond Street Hospital without postoperative complications. Postoperative assessment showed improved physical activity, the absence of noisy breathing, and easier eating. A follow-up bronchoscopy 7 months later revealed substantial improvement of the tracheal lumen. Throughout the 3.5-year follow-up, she remained symptom-free, without dyspnoea or respiratory infections, and reported improved physical performance.

Patient 5 was a male infant, born at term, identified with cystic fibrosis through newborn screening. He exhibited persistent noisy breathing and multiple episodes of BRUEs and apnoea, often triggered by feeding, starting at 4 months of age. Severe tracheomalacia, which is more prevalent in cystic fibrosis patients,[11] was confirmed by bronchoscopy, showing kissing walls throughout most of the trachea. A CT scan also revealed tracheal compression by the brachiocephalic artery. Referred to London, he underwent simultaneous TP and AP with the placement of an external Gore-Tex splint at 5 months old, with no complications. Over the 2.7-year follow-up, there was a complete resolution of severe tracheomalacia symptoms, including BRUEs, feeding difficulties and apnoea.

Patient 6 was a female infant born full term who underwent EA repair shortly after birth. She began experiencing weekly BRUEs from 1 week old and underwent AP for severe tracheomalacia at another Danish hospital at 3 months. As BRUEs persisted, often associated with meals, she was referred to Rigshospitalet, where bronchoscopy and CT confirmed severe tracheomalacia with tracheal compression by the aortic arch and a remaining thymus. At 4 months old, she underwent a second AP with partial thymectomy at Rigshospitalet, but there was no clinical improvement. This led to TP 17 days later. She was extubated 2-day post-TP but developed a central venous catheter infection, which was treated with antibiotics, allowing her discharge 7 days later. Two months after TP, bronchoscopy showed highly satisfactory tracheal conditions. During the 1.9-year follow-up, she remained clinically stable, free from BRUEs and feeding difficulties.

Patient 7 was a male born full term who underwent EA repair on his first day of life. From 4 months of age, he experienced recurrent BRUEs, frequent lung infections requiring hospital admissions, oesophageal dilation procedures, and lodged food elements causing respiratory difficulties. At 14 years old, he continued to have eating difficulties, with food frequently lodging in the oesophagus and causing airway collapse symptoms. He also reported poor physical activity performance. Bronchoscopy confirmed tracheomalacia with moderate collapse of the lower tracheal segment. He underwent TP at Rigshospitalet at age 14. His recovery was challenging, complicated by pneumothorax and atelectasis, but he was discharged 9 days later. Three months postoperatively, he experienced another instance of food lodging in the oesophagus but without respiratory difficulties. Eight months postoperatively, bronchoscopy showed reduced intrusion of the posterior wall. His spirometry flow-volume curve exhibited notable improvement, with a normalised expiratory slope. At the latest follow-up, 18 months after TP surgery, he reported ongoing improvement in physical activity and complete relief from respiratory difficulties caused by lodged food elements.

The primary aim of this study was to share our experience with the surgical management of tracheomalacia in paediatric patients, contributing valuable insights to the ongoing discussion on surgical interventions for tracheomalacia. By doing so, we also emphasise the crucial rule of national and international collaborations, particularly within the Nordic countries, to enhance and standardise tracheomalacia management.

This study describes seven cases of surgical interventions for bronchoscopy-verified severe tracheomalacia. Preoperatively, the youngest children presented with severe respiratory symptoms, including BRUEs, while those older than five primarily reported physical limitations. Overall, the interventions resulted in favourable clinical outcomes.

Previous studies have shown promising outcomes associated with surgical interventions, such as TP and AP, in managing tracheomalacia. TP has led to significant clinical improvement[7,12,13] or even resolution of respiratory symptoms, including life-threatening events[14] and a marked reduction in respiratory tract infections.[15] However, these studies had varying sample sizes (8–98 patients) and short follow-up periods. The median age at operation ranged from 2 days to 15 months. AP has also proven effective in relieving respiratory issues in patients with isolated airway compression and severe tracheomalacia.[10,16,17]

In this study, all but one patient underwent multiple operations, including both TP and AP. Three patients received simultaneous TP and AP and required no further interventions, suggesting potential benefits from combining anterior and posterior collapse prevention in certain tracheomalacia cases. This combined approach has gained international attention.[7,18] Lawlor et al.[19] reported that approximately 90% of patients with severe tracheomalacia experienced significant respiratory improvement with TP alone but adding anterior intervention for those with persistent symptoms yielded promising outcomes. Shieh et al.[14] found that eight patients who underwent AP before TP, and 11.2% of their TP patients, required additional AP. This combined approach may hold relevance in severe cases, warranting further research to determine the optimal surgical strategy.

Managing tracheomalacia requires a comprehensive approach beyond selecting surgical interventions. While addressing acute life-threatening events is crucial, it is equally important to manage the full spectrum of tracheomalacia-related symptoms, including the physical limitations observed in our two older patients. Interestingly, our centre primarily pursued surgical intervention as a last resort for critically ill younger patients with life-threatening events, differing from the more preventive approaches adopted by other centres. For instance, centres in Nagoya, Japan,[13] Utrecht, Netherlands[4] and Boston, USA,[20] incorporated TP during primary EA repair if significant tracheomalacia was diagnosed perioperatively. Nagoya[13] reported no additional complications with TP during EA repair, and Boston[20] found a significant reduction in respiratory morbidity within the first year of life. In a comparison by Shieh et al.[21] outcomes

and risks were similar whether TP was performed during EA repair for 18 patients or as a secondary treatment for 100 patients. Given the frequent association between EA and tracheomalacia, it is necessary to evaluate the potential for combining surgical interventions during EA repair, as this may help avoid subsequent operations and prevent respiratory complications.

Variations in the timing and criteria for surgical intervention across countries have sparked debate about our team's current last-resort approach. Expanding surgical indications could potentially prevent clinical deterioration in severe tracheomalacia cases and address milder pulmonary manifestations. However, significant gaps remain in understanding the long-term outcomes of tracheomalacia surgeries.[4,12,13,20] Furthermore, these procedures require refined surgical techniques.[10,20,22] Therefore, a thorough evaluation of the risks and benefits of adopting a more preventative approach is essential for this population. International collaboration and data collection are crucial to address these uncertainties.

We strongly encourage all centres to share their experience in managing tracheomalacia and consider collaborating on an international registry to enhance future research efforts. While our institution, Rigshospitalet, has primarily collaborated with Great Ormond Street Hospital, there is considerable potential for both national and international partnerships, particularly within Nordic countries.

In conclusion, our case series provides valuable insight into the surgical management of tracheomalacia in children, emphasising the need for a standardised intervention protocol that carefully considers potential benefits and risks. We advocate for enhanced national and international collaboration to increase patient volume and surgical expertise, promoting a multidisciplinary approach to optimise patient care. Both AP and TP, whether performed individually or in combination, have shown promising results for severe tracheomalacia, with favourable outcomes observed during follow-up. Early intervention appears particularly beneficial, underscoring the importance of timely treatment. However, current experience and data remain limited, necessitating a larger dataset with diverse patient subgroups. Future research should prioritise clarifying long-term outcomes and refining patient selection criteria to optimise treatment efficacy and safety. Ideally, these efforts should be conducted throughout multicentre studies to provide comprehensive insights and robust evidence.

AUTHOR CONTRIBUTIONS

Helene Holmark Møller: Writing – original draft; methodology; investigation; data curation; formal analysis; project administration. **Marika Nathalie Schmidt:** Writing – review and editing; conceptualization; methodology; formal analysis. **Inge B. R. Ifaoui:** Writing – review and editing. **Hans G. H. Thyregod:** Writing – review and editing. **Elisabeth S. Christiansen:** Writing – review and editing. **Nagarajan Muthialu:** Writing – review and editing. **Frederik F. Buchvald:** Writing – review and editing. **Kim G. Nielsen:** Writing – review and editing; supervision.

FUNDING INFORMATION

This study did not receive any specific funding.

CONFLICT OF INTEREST STATEMENT

The authors have no conflicts of interest to declare.

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How to cite this article: Møller HH, Schmidt MN, Ifaoui IBR, Thyregod HGH, Christiansen ES, Muthialu N, et al. Tracheomalacia and surgical options: A case series perspective. Acta Paediatr. 2025;114:59–64. https://doi.org/10.1111/apa.17452