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Thoracoscopic Stage Internal Traction Repair Reduces Time to Achieve Esophageal Continuity in Long Gap Esophageal Atresia

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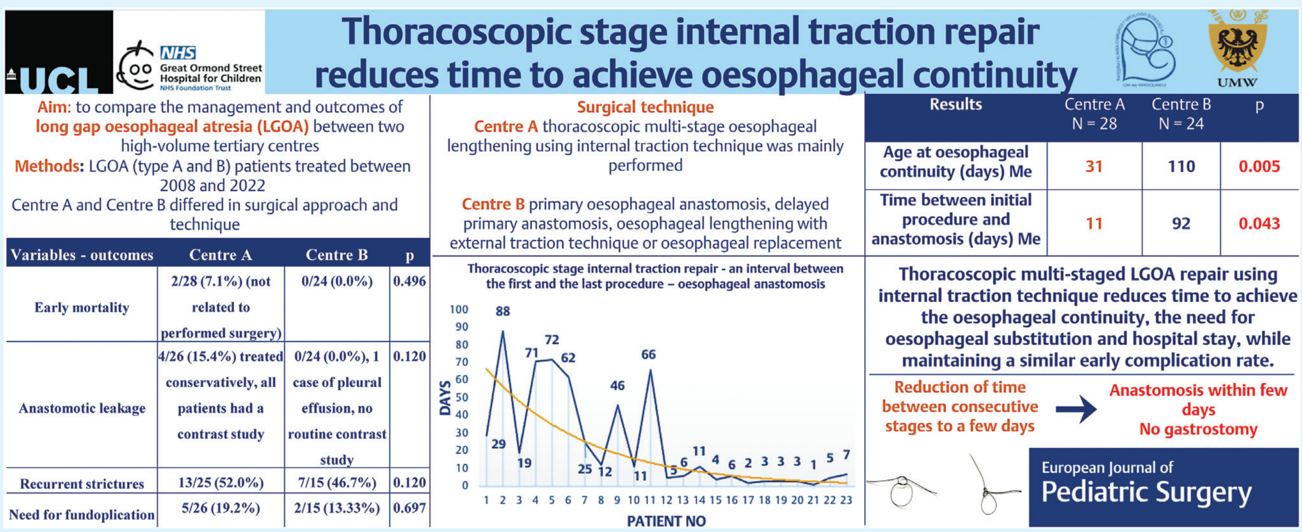
Graphical Abstract



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Abstract

Objective Management of long gap esophageal atresia (LGOA) is controversial. This study aims at comparing the management of LGOA between two high-volume centers.
Methods We included patients with LGOA (type A and B) between 2008 and 2022. Demographics, surgical methods, and outcomes were collected and compared.
Results The study population involved 28 patients in center A and 24 patients in center B. A surgical approach was thoracoscopic in center A, only for one patient was open for final procedure. In center B, 3 patients were treated only thoracoscopically, 2 converted to open, and 19 as open surgery. In center A primary esophageal anastomosis concerned 1 case, two-staged esophageal lengthening using external traction 1 patient, and 26 were treated with the multistaged internal traction technique. In 24 patients a full anastomosis was achieved: in 23 patients only the internal traction technique was used, while 1 patient required open Collis–Nissen procedure as final management. In center B primary anastomosis was performed in 7 patients, delayed esophageal anastomosis in 8 patients, esophageal lengthening using external traction in 1 case, and 9 infants required esophageal replacement with gastric tube. Analyzed postoperative complications included: early mortality, 2/28 due to accompanied malformations (center A) and 0/24 (center B); anastomotic leakage, 4/26 (center A) treated conservatively—all patients had a contrast study—and 0/24 (center B), 1 case of pleural effusion, but no routine contrast study; recurrent strictures, 13/26 (center A) and 7/15 (center B); and need for fundoplication, 5/26 (center A) and 2/15 (center B). Age at esophageal continuity was as a median of 31 days in center A and 110 days in center B. Median time between initial procedure and esophageal anastomosis was 11 days in center A and 92 days in center B.
Conclusion Thoracoscopic internal traction technique reduces time to achieve esophageal continuity and the need for esophageal substitution while maintaining a similar early complication rate.

Keywords
- long gap esophageal atresia
- esophageal atresia
- internal traction
- minimally invasive surgery
- delayed primary anastomosis

Introduction

Long gap esophageal atresia (LGOA) represents approximately less than 10% of all OA cases and is a complex malformation, often associated with other defects.[1] The management of LGOA remains controversial and challenging. It is further challenged by diversity in case definition, limited numbers of cases, a variety of surgical techniques used, as well as insufficient guidelines for best practice.[1–6] Determination of the optimal management therefore requires collaboration and cooperation between experienced centers. In 2019, the European Reference Network for Rare Inherited Congenital Anomalies (ERNICA) conducted the first consensus study on the treatment and follow-up of LGOA patients.[3] Complete agreement was achieved on the statements of preferential preservation of the native esophagus with delayed primary anastomosis (DPA), as well as centralization of treatment of LGOA in expert centers.[1,3] Consensus also was reached to determine thoracoscopic esophageal mobilization and lengthening using traction sutures as a promising method in experienced departments.[1,3]

There are many definitions of LGOA, including: type of OA (i.e., type A + B), the length of the gap between upper and lower pouches (measured in vertebral bodies or as an absolute measurement), or the surgeon's inability to perform an anastomosis.[1–3,7,8] Koivusalo et al included type C anomalies with tracheoesophageal fistula (TEF) located at the carina or below as LGOA.[9] The International Network of Esophageal Atresia working group, as well as ERNICA consensus indicated LGOA as any esophageal atresia without air in the abdomen.[2,3]

The aim of the study is to compare management and outcomes of LGOA patients between two high-volume referral centers with expertise in esophageal atresia and newborn surgery. Both departments differed considerably in surgical approach, repair techniques, and definition of LGOA.[1,10]

Materials and Methods

Patients at center A underwent LGOA repair at the Department of Pediatric Surgery and Urology of University Clinical Hospital in Wroclaw in Poland. Those at center B were operated on at the Department of Specialist Neonatal and Pediatric Surgery of Great Ormond Street Hospital for Children in London in the United Kingdom. Patients treated over a 15-year period between January 2008 and December 2022 were included. This study received local ethical approval in both sites (Ethical Committee of Medical University in Wroclaw: 169/2022, 24.02.2022; and Audit 2919 at Great Ormond Street Hospital).

Centers differed in their standard definition of LGOA: in center A all type A and B were defined as LGOA. Comparatively, in center B a gap length precluding primary anastomosis defined LGOA cases. In the study we defined LGOA using the subjective measure of anomaly type, and therefore all cases of type A and type B were included in the subsequent analysis.

The clinical data of included patients were retrospectively collected from medical records. Time between initial procedure and the esophageal anastomosis, age at esophageal continuity, as well as time between consecutive stages among multistage repairs were obtained and analyzed. The surgical details included initial management and approach, definitive surgery, conversion rate, and other interventions connected to esophageal repair such as number of endoscopic dilatations and recurrent strictures. Postoperative complications were analyzed and included: early mortality, anastomotic leakage, recurrent esophageal strictures, and need for antireflux surgery. Recurrent esophageal strictures were considered as more than symptomatic esophageal obstruction, requiring more than one endoscopic dilatation. Neither center performed elective dilatations in patients without symptoms. Early mortality was defined as mortality before discharge. The causes of early death were determined to be related to surgery or to result from patient factors. Patients have been followed up routinely in the outpatient setting with additional studies (e.g., contrast imaging), if clinically indicated.

In both centers, the surgical treatment started with a preoperative bronchoscopy to identify a potential proximal TEF and possible any malformations of trachea and larynx including vocal cord palsy, tracheomalacia, and laryngeal cleft. Standard practice at center A is to utilize a thoracoscopic approach as a procedure of choice. For cases of LGOA, a thoracoscopic multistage esophageal lengthening procedure is preferred using internal traction sutures.[4,10] It is feasible to carry the procedure out within a few days after birth, even avoiding a gastrostomy placement in certain cases where the stages of the procedure can be performed over a short period of time.[4,10] The internal traction suture technique was based on two sliding knots which allowed to approximate the esophageal ends with constant traction force, dispersed among clips placed across the esophageal tips.[4,10] During the next stage procedure, the sliding knots were reconfigured to get the pouches closer or if both the pouches overlapped each other, an anastomosis would have been started.[4,10] The anastomosis was always performed over a nasogastric (NG) tube, which was left until a postoperative contrast study, usually on the 5th to 6th day after surgery. A chest cavity drain was placed in some cases, especially with difficult anastomosis under high tension. Patients between stages stayed intubated at the intensive care unit, on total parenteral nutrition and intermittent, oral suction as needed was applied.

In center B only 3/24 patients were treated from birth, while the remainder of the cohort studied were referred from other centers in the region after diagnosis as LGOA. Ultimate surgical management involved different techniques: primary esophageal anastomosis, DPA, esophageal lengthening using external traction technique, or esophageal replacement with gastric tube. Use of thoracotomy or thoracoscopy was defined by surgeon preference. The strategy involved esophageal mobilization, an initial gap assessment, followed by the decision to perform primary anastomosis if possible. If not, a gastrostomy was placed, and the definitive management was postponed until approximately 3 months of age to reevaluate the feasibility of the DPA or to proceed to esophageal replacement, mainly with a gastric tube. Gap assessment between subsequent stages was performed with fluoroscopy to check the change of distance and to estimate a proper time for the next procedure. While awaiting esophageal continuity to be achieved, patients remained inpatient at the hospital.

Postoperative contrast study was performed in all patients from center A to confirm proper esophageal passage and exclude any anastomotic leakage, then oral feeding was started, and the NG tube was removed if swallowing was undisturbed. At center B, there was no routine contrast study; however, a chest drain was used to identify and monitor any anastomotic leakage.

Verification of the normality of quantitative variables was performed with the Shapiro–Wilk's test. Qualitative variables were reported as mean values $\pm$ standard deviation or, if not normally distributed, as median and interquartile range, while categorical variables were reported as numbers (n) and percentages (%). Quantitative variables were compared using unpaired t-test or Mann–Whitney U test (for nonparametric variables), while categorical variables were compared using chi-square test or Fisher's exact test. A p-value of less than 0.05 was considered statistically significant.

Results

Characteristics of Study Population and Surgical Treatment

In the period of study, 28 patients with type A and B anomalies were treated at center A, compared with 24 at center B. Patient characteristics are summarized in ►Table 1. Of note, 25/28 (89%) patients received their care in center A from birth compared with only 3/24 (13%) patients underwent the first procedure in center B. At center A, all patients were treated by a single surgeon, while at center B, six consultants were involved in the care of LGOA patients. Twelve patients from center A and 10 patients from center B were treated without gastrostomy.

Time to Restore Esophageal Continuity and between Consecutive Stages

Out of 28 patients with LGOA in center A, 1 primary anastomosis was performed, 1 patient was repaired using thoracoscopic Foker technique with external traction, and 26 patients were treated with the internal traction technique described in the "Methods" section. There was no conversion to open surgery at initial management. In 26 patients a full anastomosis was performed: in 25 patients only by thoracoscopic approach and 1 patient required esophageal replacement due to upper pouch perforation by the traction suture. The patient had a cervical esophagostomy, thoracoscopic distal pouch lengthening, and finally an open Collis–Nissen procedure. Three patients died, all due to associated malformations (2 cases of early death, before anastomosis, and 1 case of late mortality, who had a completed anastomosis and

Table 1 Characteristics of study population and operative treatment

Variables	Center A ($N = 28$)	Center B ($N = 24$)	p-Value
Gender			
Female, n (%)	13 (46.4)	12 (50.0)	1.000
Male, n (%)	15 (53.6)	12 (50.0)	
Gestational age (completed weeks)			
Mean ± SD	35.6 ± 2.5	35.2 ± 3.5	0.634
Median [Q1; Q3]	36 [35; 37]	36 [34; 37.25]	
Min—Max	29–39	26–40	
Birth weight (g)			
Median [Q1; Q3]	2,500 [2,190; 2,605]	24,00 [2,035; 2,578]	0.812
Min–Max	750–3,200	940–4,475	
Type of OA (gross classification)			
A	12 (42.9)	18 (75.0)	**0.026**
B	16 (57.1)	6 (25.0)	
First approach			
Thoracoscopic, n (%)	28 (100)	5 (20.8)	< **0.001**
Open, n (%)	0 (0.0)	19 (79.2)	
Conversion rate, n (%)	0/28 (0.0)	2/5 (40.0)	
Initial management			**0.019**
First surgery at the treatment center	25/28 (89.3)	3/24 (12.5)	
Primary esophageal anastomosis, n (%)	1 (3.5)	7 (29.2)	**0.010**
Esophageal lengthening, n (%)	27 (96.5)	1 (4.2)	
Internal traction, n (%)	26 (93.0)	0 (0.0)	
External traction, n (%)	1 (3.5)	1 (4.2)	
Gap assessment alone, n (%)	0 (0.0)	10 (41.7)	
Gap assessment + esophagostomy, n (%)	0 (0.0)	6 (25.0)	
Gastrostomy, n (%)	16 (57.1)	14 (58.3)	0.930
Definitive management			
Delayed esophageal anastomosis, n (%)	24 (85.7)	8 (33.3)	< **0.001**
Esophageal replacement, gastric tube, n (%)	1 (3.5) (Collis–Nissen procedure at the final stage)	9 (37.5)	**0.002**

Abbreviations: OA, esophageal atresia; SD, standard deviation. Bold values signify p-value < 0.05.

functioning esophagus). The average time interval between first and second procedure of thoracoscopic staged OA repair using internal traction technique for the first 9 patients was 28.7 days (median 29) and it significantly was decreased for the last 14 patients to mean value of 3.4 (median 3). Twelve patients were operated on without gastrostomy.

Concerning the 24 patients at center B, primary esophageal anastomosis was performed in 7 patients, and 8 patients were treated as delayed esophageal anastomosis (in 1 of them using external traction technique). Nine patients required esophageal replacement, the majority with a gastric tube. In all cases, an esophageal continuity was accomplished. The initial approach was via thoracotomy in 19 patients and thoracoscopy in 5 patients with a conversion to open access in 2/3.

While the number of surgical stages was higher, the number of days needed to achieve esophageal continuity was significantly shorter in center A (►Table 2 and ►Fig. 1).

Complications

Postoperative complications from both centers are depicted in ►Table 3. There was no statistical difference between centers for early mortality, anastomotic leakage, recurrent stricture, or eventual need for antireflux surgery.

Discussion

The main goal of this study was to compare management and outcomes of surgical LGOA repair between two high-volume tertiary centers from different European countries who have

Table 2 Number of stages of surgical repair, age at esophageal continuity, and intervals between initial and definitive management

Variables	Center A (N = 28)		Center B (N = 24)	p-Value
	Completed anastomosis in total, N = 26 (2 patients died before anastomosis)	Completed anastomosis only with internal traction, N = 23		
Number of stages/ surgical procedures to achieve continuity				––
Mean ± SD	2.9 ± 1.7	2.7 ± 1.1	2.2 ± 1.2	
Median [Q1; Q3]	2 [2; 3]	2 [2; 3]	2 [1; 3]	
Min–Max	1–9[a]	2–6	1–6	
Age at esophageal continuity (d), median [Q1; Q3]	31 [9.75; 72.25]	30 [9; 72]	110 [58–317]	**0.005**
Days between initial procedure and anastomosis, median [Q1; Q3]	11 [3.75; 53.75]	11 [4; 46]	92 [2–324]	**0.043**

Abbreviation: SD, standard deviation.
[a]One patient with complicated postoperative course—upper pouch perforation after second thoracoscopy, emergency esophagostomy, left-side thoracoscopy—mobilization and elongation of lower pouch, final Collis–Nissen open repair.

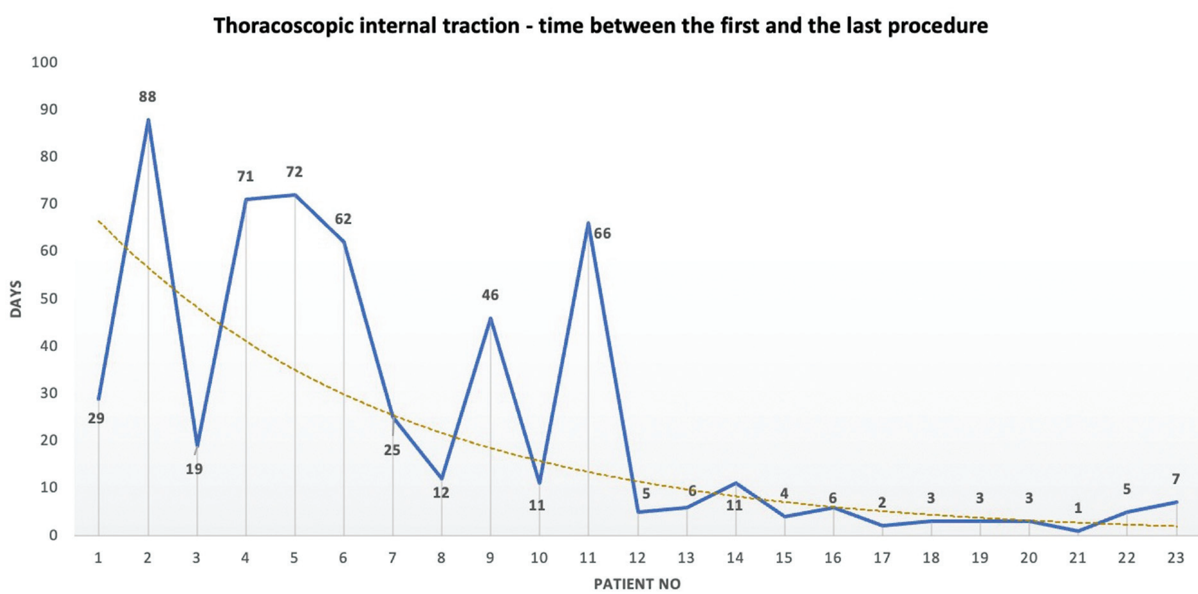

Fig. 1 Illustrated the interval between the first and the last procedure—esophageal anastomosis.

adopted a different approach to managing this condition. The main findings refer to differences related to the numbers of surgical procedures, the number of esophagi which were able to be preserved, and time to reach final anastomosis/ substitution. As there are widely varied definitions of LGOA worldwide, including among our two centers, we included only type A and B anomalies to assure appropriate comparisons could be made.

Current consensus is that preservation of the native esophagus is preferred when possible, because no substitute can effectively replace its function.[1–3,11–13] In LGOA, esophageal anastomosis can be particularly challenging and DPA has been the preference in center B. This management might be feasible and effective even in cases with the distance more than 3.5 cm.[1–3,12,14–17] However, this approach clearly has disadvantages while waiting for the esophageal spontaneous growth such as need for gastrostomy placement, prolonged hospital stays (with associated impact on the wider family unit), and the constant threat of aspiration pneumonia requiring regular upper pouch suction.[15] Moreover, DPA has been associated with high occurrence of gastroesophageal reflux (GER),[13,14] although the need for fundoplication

Table 3 Postoperative complications

Variables	Center A ($N = 28$)	Center B ($N = 24$)	*p*-Value	OR
Early mortality	2/28 (7.1%)	0/24 (0.0%)	0.496	4.30 (0.20–93.9)
Anastomotic leakage	4/26 (15.4%), treated conservatively with chest drainage, all patients had a contrast study	0/24 (0.0%), 1 case of pleural effusion, no routine contrast study	0.120	8.32 (0.43–163)
Recurrent strictures	13/25 (52.0%)	7/15 (46.7%)	0.120	1.11 (0.36–3.41)
Need for fundoplication	5/26 (19.2%)	2/15 (13.33%)	0.697	1.44 (0.31–6.54)

Abbreviation: OR, odds ratio.
Note: In center A $N = 25$ and in center B $N = 15$ for recurrent strictures after esophago-esophageal anastomosis, cases of esophageal replacement were excluded due to different mechanism and anatomy of stricture formation.

was in fact lower in the patients who retained their native esophagus at center B than in center A.

Esophageal substitution has been advocated in case of failure of DPA or in cases of a very wide gap, when anastomosis is not attainable.[18] In center B, a gastric interposition was performed in 37.5% patients, while only 1 patient from center A required a Collis–Nissen as the final management due to complications of the primary repair. Esophageal replacement with gastric tube revealed similar perioperative risk, but more long-term complications than DPA and 90% of the whole population regardless of type of surgical repair had GER.[8]

Esophageal elongation techniques have been developed in the last two decades and harness the growth potential of a newborn esophagus to shorten the time to esophageal continuity. Foker at al established a growth-induction technique based on placement of external traction sutures on esophageal tips, which were approximated toward each other with a tension being increased by retightening of suture knots.[19] This technique was performed as open surgery; however, a thoracoscopic LGOA repair using external traction sutures was first performed by van der Zee et al in 2007.[20,21] It is out of this article's scope whether it is growth or distension induced by the traction of esophageal pouches.

Based on Foker's initial concept and using an minimally invasive surgery (MIS) approach, Patkowski has developed a thoracoscopic multistaged LGOA repair using internal traction sutures.[4,10] Initially, the time between consecutive stages was intentionally 3 to 5 weeks and most cases were repaired in two stages.[10,22] However, patients treated with shorter intervals did not affect the chance to achieve a successful two-staged anastomosis.[22] Based on these findings and the similar experience of van der Zee et al,[21] a reduced time between consecutive stages to a few days has allowed to perform the anastomosis within the 2 weeks life and avoid the gastrostomy placement in most recent cases.[10,22] Assessing the most recent 9 patients, the thoracoscopic internal traction technique requires a median of 5 days to achieve esophageal continuity (range 1–11 days). This compares to a median time from initial procedure to anastomosis of 92 days at center B, which was of course statistically significant and bears considerable psychological impact on the parents and siblings of and affected child as well as associated inpatient health care costs. Similarly, a large multicenter study of management and outcomes of isolated LGOA treatment using DPA revealed a high rate of successful reconstructions, but prolonged length of hospital stay (median 125 days) with a median age at repair of 87 days.[23] A long initial hospital stay, as well as significant associated anomalies, and persistent digestive or respiratory symptoms may negatively influence on health-related quality of life.[24–26] Other studies have also remarked that a prolonged hospital stay may be related to suboptimal long-term morbidity and neurodevelopment outcomes.[27,28]

Postoperative complications such as gastroesophageal reflux disease with need for antireflux surgery and anastomotic strictures were similar among analyzed centers. Although not statistically significant, there were observed differences in the rate of anastomotic leakage (15% for center A and 0% for center B), which may result from the utilization of routine postoperative contrast study; importantly, there were no leaks that required surgical management. One patient from center B had a pleural effusion and was also treated conservatively. In both departments there were no mortality related to surgery, and the fact that the majority of patients at center B were referred from other centers for ongoing operative management may prejudice the cohort to remove patients who had unsurvivable associated anomalies (vs. 3 patients in center A).

Centers differed significantly in their use of MIS. In our study the approach was thoracoscopic in all cases from center A compared with 5 newborns from center B, with a conversion rate of 40%. Thoracoscopy, which allows repeated procedures in the chest with minimal harm, may contribute to quicker recovery, diminished pain, and has been shown to reduce late thoracic musculoskeletal morbidity.[4,6,10,29–32] MIS also has the value of allowing precise assessment of mediastinal anatomy, the quality of the tissue of the esophageal pouches, and even perfusion using advanced technologies with minimal risks for patient.[4,33] Moreover, MIS techniques allow to perform the consecutive stages of OA repair every few days with less systemic inflammatory response and fewer adhesions, characteristic for the open approach.[6,10,21] Reports from centers across the United

Kingdom of esophageal lengthening with traction have described a high rate of complications (including esophageal pouch leak or disruption) and an associated prolonged hospital stay; however, it should be noted that these reported outcomes are based on assessment of cases from many departments with comparatively few cases at each; reinforcing the ERNICA consensus that these complicated cases should be concentrated into high-volume centers.[34,35]

The study is limited by the retrospective nature of the comparison, and several aspects of the data presented were not assessed by comparable means (i.e., postoperative anastomotic leak). Moreover, while all LGOA cases from center A were repaired by one single surgeon, in center B 24 patients were treated by six surgeons with the obvious associated variability of practice. We would suggest that performing a larger number of cases by a limited number of surgeons may help when introducing and refining a new technique.[4,5] Finally, it is difficult to make definitive conclusions regarding long-term outcomes since follow-up periods varied from 6 months to 14 years and there has not been an evaluation of the functional outcomes of quality of life of these children. Despite these limitations, this is the comparison of two successful strategies of LGOA management, emphasizing the positive outcomes of esophageal continuity with minimal morbidity that can be achieved in high-volume centers. Although the preservation of the native esophagus is preferable, some cases may require substitution. In the future, esophageal tissue engineering may overcome the need for substitution and therefore offers a real advantage for the treatment of LGOA patients.[36,37]

Conclusion

The surgical management of LGOA patients remains controversial and all currently accepted options are technically challenging. Despite differences between the two centers in this study, the postoperative complications were similarly infrequent. When performed in specialist centers, thoracoscopic multistaged LGOA repair using internal traction technique can be shown to reduce time to achieve esophageal continuity, resulting in reduced hospital stay and the need for esophageal substitution, while maintaining a similar early complication rate. There is also a potential lower long-term morbidity resulting from consecutive use of minimally invasive technique, but it requires further follow-up and studies. Due to the low number of cases with LGOA we feel these results reinforce the need to push for centralization of care, as well as cooperation between institutions to improve the outlook for children born with LGOA.[38]

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Conflict of Interest

None declared.

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