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Childhood outcome after correction of long-gap esophageal atresia by thoracoscopic external traction technique

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ABSTRACT

Background: Thoracoscopic external traction technique (TTT) is a relatively new surgical intervention for patients with long-gap esophageal atresia (LGEA) that preserves the native esophagus. The major accomplishment with TTT is that esophageal repair can be achieved within days after birth.
This study evaluates the childhood outcome in LGEA patients treated with TTT, including gastrointestinal outcome, nutritional status and Health-Related Quality of Life (HRQoL).
Methods: A cohort study including all LGEA patients that underwent TTT between 2006-2017 was conducted. Patients and/or their parents were invited to fill out questionnaires regarding reflux symptoms and HRQoL.
Results: TTT was successful in 11/13 patients (85%). Esophageal anastomosis was accomplished at a median age of 12 days (range 7-138), first oral feeding was started at a median of 16 days postoperatively (range 5-37). All patients required multiple dilatations and 10 patients required anti-reflux surgery.
At median follow-up of seven years, five patients reported mild and one moderate reflux complaints. All patients but one reached age-appropriate oral diet. Most patients (80%) were within normal growth range. Overall HRQoL was comparable to healthy controls.
Conclusion: TTT provides acceptable results in childhood. Oral feeding can be started as soon as two weeks postoperatively. Almost all patients are able to eat an age-appropriate oral diet. Overall HRQoL was comparable to healthy controls.

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Introduction

Long-gap esophageal atresia (LGEA) is a rare and complex type of esophageal atresia (EA) and accounts for approximately 10% of all newborns with EA [1–3]. In literature the definition of LGEA is inconsistent. Recently, however, the International Network of Esophageal Atresia (INoEA) has defined LGEA as "any esophageal atresia without abdominal air", corresponding to Gross type A and B [4].

Bridging the wide gap in LGEA remains a challenge for pediatric surgeons [5] and several surgical approaches have been described. Esophageal repair can be performed by esophageal replacement (e.g. jejunal or colon interposition or by gastric pull-up [6–8]). However, most surgeons agree that the native esophagus is the best esophagus [9]. Preservation of the native esophagus can be accomplished by delayed primary anastomosis or by open or thoracoscopic traction technique [10,11]. Delayed primary anastomosis entails that the esophagus is restored two to three months after birth [12]. Prolonged delay of esophageal continuity may lead to several disadvantages, including swallowing difficulties due to postponed oral feeding and prolonged hospital stay [13–15]. With the thoracoscopic external traction technique (TTT) however, as also developed by our center, esophageal repair can be accomplished within days after birth [5,11]. Although several studies have been conducted on the outcome after esophageal

Abbreviations: DPA, delayed primary anastomosis; EA, esophageal atresia; FOIS, functional oral intake scale; GER, gastroesophageal reflux; HRQoL, Health-Related Quality of Life; LGEA, long-gap esophageal atresia; TTT, thoracoscopic external traction technique.

LEVEL OF EVIDENCE: IV
Type of study: Prognosis study

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replacement and delayed primary anastomosis in LGEA, this is the first study that evaluates the childhood outcome after the TTT.

The aim of this study is to evaluate the outcome in childhood in LGEA patients treated with TTT, including gastrointestinal outcome, nutritional status and Health-Related Quality of Life (HRQoL).

Methods

Study design and participants

A retrospective cohort study was conducted including all LGEA patients corrected via the thoracoscopic external traction technique (TTT) at the University Medical Center Utrecht, Wilhelmina Children's Hospital, between 2006 and 2017. Electronic medical records were reviewed. All LGEA patients since 2006 were treated with TTT and patients were considered eligible for the study if TTT for repair of LGEA was completed. Only patients with Gross type A or B (no distal tracheoesophageal fistula) were included. Patients with a failed procedure were excluded from further analysis. Patients with Down syndrome were excluded from analysis of questionnaires.

Surgical procedure

Prior to surgery, a standard rigid bronchoscopy was performed in almost all EA patients to evaluate the presence of a proximal fistula and to evaluate the severity of tracheomalacia. The subsequent TTT has previously been described by Van der Zee et al., [5,11]. In short, thoracoscopic traction sutures were placed at both esophageal ends and were fixed externally with mosquito forceps. Approximation of the esophageal ends was evaluated by postoperative X-rays. When this approximation hampered prematurely, thoracoscopic adhesiolysis was performed. Both ends were anastomosed during a final thoracoscopic procedure. A chest tube was positioned next to the esophageal anastomosis. Initially a gastrostomy was placed for feeding, in later patients a laparoscopic gastropexy was performed to prevent the stomach from migrating into the thorax. Patients were kept on parenteral feeding during the traction period.

Clinical assessment

Baseline characteristics, including gender, gestational age, birthweight, type EA and associated anomalies were collected from the medical records. All patients had regular check-ups at the Wilhelmina Children's Hospital and since 2017 a multidisciplinary routine follow-up schedule (age 0 up to 17 years) had been introduced for all EA patients. Gastrointestinal and respiratory symptoms, development and health-related-quality of life parameters were assessed.

Surgical outcome

Surgical data, including age at surgery, traction time and gastrostomy or gastropexy placement were obtained. Postoperative data, including ventilation time, NICU and hospital length of stay, postoperative complications (i.e. leakage, stenosis) and first enteral and oral feeding were collected.

Gastroesophageal reflux

Validated reflux-questionnaires were used to define gastroesophageal reflux (GER). Two different questionnaires were used for evaluation of GER and dysphagia. The age-adjusted Gastroesophageal Reflux Symptom Questionnaire (GSQ) [16] was used for patients from 2 to 12 years old and the Reflux Disease Questionnaire (RDQ) [17,18] was used for children of 12 years and older. The GSQ-questionnaire was available as parent-proxy report and contains questions on the frequency (n) and severity of reflux and dysphagia in the past seven days, which was scored for severity on a 7-point Likert scale ranging from 1 (none) to 7 (most severe). The RDQ was available as self-report and contains questions on the frequency and severity of regurgitation, heartburn and dyspepsia in the past seven days, which were scored on a 6-point Likert scale from 0 (never/none) to 5 (daily/most severe). Symptoms for all questionnaires were divided in four categories: no symptoms, mild (mild symptoms weekly), moderate (mild symptoms daily or severe symptoms weekly) and severe symptoms (severe symptoms daily).

Functional oral intake scale (FOIS)

The functional oral intake scale (FOIS) was used to evaluate oral intake. It consists of a numeric scale concerning oral intake, ranging from 1 (nothing by mouth) to 7 (full oral diet, no restrictions) [19].

Nutritional status

Weight and height measurements were collected and converted into the weight-for-length z-score using the Netherlands Organization for Applied Scientific Research (TNO) growth standards [20]. A *z*-score below -2SD was considered pathological [21,22].

Health-related quality of life (HRQoL)

Health-Related Quality of Life was evaluated using the age-adjusted Pediatric Quality of Life Inventory (PedsQL^{TM}) 4.0 Generic Core Scales questionnaire. Patients and/or their parents were asked to fill out this questionnaire. It encompasses the domains physical functioning, emotional functioning, social functioning and (pre-)school functioning and it was scored for frequency on a 5-point Likert scale from 0 (never) to 4 (almost always). Scores were transformed to a 0-100 scale, with a higher score representing a better HRQoL. Scores were compared to healthy controls with a total scale cut-off point score of -1SD below the population sample mean (69.7 for child self-report and 65.4 for parent-proxy report). Scores of 1 SD below the mean of the healthy population are at risk for an impaired HRQoL [23].

Statistical analysis

Nonparametric variables are presented as median and range and categorical data is presented as frequencies and percentages. Mean differences are presented with 95% confidence intervals. Data from children and their parents was treated as paired.

The analyses were performed with SPSS for Windows, version 25.0 (IBM Corp., Armonk, NY) and R 4.0.0 (R Core Team, Auckland, New Zealand).

Ethical approval

This cohort study was submitted to the UMCU Ethics Committee. No ethical approval was required according to the Medical Research Involving Human Subject Act. The study was carried out in accordance with the Declaration of Helsinki. Informed consent from all patients, and/or their parent if applicable, was obtained before sending the questionnaires.

Results

Between 2006 and 2017, a total of 14 patients with long-gap esophageal atresia were operated in the Wilhelmina Children's Hospital. Three patients were excluded from further analysis: a primary thoracoscopic repair was feasible in one patient and the elongation procedure failed in two patients. Of the two failed patients, the traction sutures tore down in the first patient (type A) and a subsequent jejunal position was performed. The length of

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Table 1
Patient characteristics.

Variable	n = 11
Male (n, %)	5 (46%)
Gestational age (weeks) (median, range)	34^{+4} (range 30^{+2} – 39^{+6})
Birthweight (g) (median, range)	1915 (range 1360–3643)
Apgar score (median, range)	
1 min	7 (range 2–9)
5 min	8.5 (range 5–9)
Gross type EA (n,%)	
Type A	6 (55%)
Type B	5 (46%)
Associated anomalies (n,%)	8 (73%)
Down's syndrome	1 (9%)
VACTERL	1 (9%)
ARM	1 (9%)
Renal	1 (9%)
Musculoskeletal	3 (27%)
Cardiac	2 (18%)
Other	3 (27%)
Tracheomalacia (n,%)	6 (55%)

EA=esophageal atresia; ARM=anorectal malformations; VACTERL=Vertebral defects, Anal atresia, Cardiac defects, Trachea-Esophageal malformation, Renal anomalies and Limb abnormalities.
Other: microcephaly, hemangiomas, microtia, retrognathia, hearing loss

hospital stay was 44 days. There was no sign of leakage and the patient required no fundoplication. In the second patient (type B), the proximal pouch was perforated by the Replogle tube and a gastric pull-up was performed. This patient was premature (33 weeks) and had a concomitant anorectal malformation. The length of hospital stay was 133 days. Since TTT failed in these patients, they were not evaluated in the analysis of the follow-up. All subsequent TTT procedures since 2013 were successful.

TTT could be completed in eleven patients, of which five (46%) were male. The median gestational age was 34^{+4} weeks (range 30^{+2}-39^{+6}) with a median birthweight of 1915 grams (range 1360-3643). Five patients (46%) had a proximal fistula (type B) and six patients (54%) had EA type A. Eight patients (73%) had associated anomalies (e.g. musculoskeletal, cardiac). Patient characteristics are presented in **Table 1**.

Surgical outcome

Traction sutures were placed at a median age of 9 days (range 2–134) and esophageal anastomosis was accomplished at a median age of 12 days (range 7–138). A definitive reconstruction was performed with two thoracoscopic procedures in four patients and with three procedures in seven patients. Four patients were transferred from either another Dutch hospital or from abroad (the child that was operated at the age of 134 days). In these four patients that were transferred from another hospital a gastrostomy had been placed before referral. At the Wilhelmina Children's Hospital only the first patient had a laparoscopic gastrostomy. In the other six patients, a TTT was performed without a gastrostomy. In four out of these six patients a laparoscopic gastropexy was performed when the traction sutures were placed, to prevent the stomach from sliding up into the chest. In the other two patients neither a gastropexy nor a gastrostomy was performed. The first patient developed a partial migration of the stomach into the thorax and a subsequent laparoscopic fundoplication was performed after 9 weeks. In the second patient, there was no tension on the distal esophageal pouch and therefore a gastropexy was not indicated.

Postoperative outcome

After final surgery, in which the esophagus was successfully anastomosed, patients remained at the NICU for a median time of 18 days (range 3–37) with a median ventilation time of four days (range 2-14). The median initial hospital length of stay was 47 days (range 27–170). The patient who had been admitted for 170 days, suffered from respiratory incidents due to severe tracheomalacia and needed an aortopexy and redo aortopexy. During his hospital stay, the patient also required multiple anastomotic dilatations, a fundoplication and a redo fundoplication. Postoperative leakage occurred in 5 patients (46%). All leakages were treated conservatively with chest tubes and antibiotics. Surgical outcome data is presented in **Table 2**.

Table 2
Surgical data.

Variable*(days)*	**n = 11***(median, range)*
Age at first surgery for EA	9 (2–134)
Age at final anastomosis	12 (7–138)
Traction days	4 (2–10)
Postoperative ventilation time	4 (2–14)
Postoperative ICU stay	18 (3–37)
LOS	47 (27–170)

EA=esophageal atresia; ICU=intensive care unit; LOS=length of hospital stay (for esophageal repair)

Table 3
Gastrointestinal outcome.

Variable	n = 11
First enteral feeding (postoperative)	3 (0-21) days
No-gastrostomy	3 (1–7) days
Gastrostomy	3 (0–21) days
Full enteral feeding (postoperative)	10 (0–27) days
No-gastrostomy	10 (5–14) days
Gastrostomy	11 (0–27) days
First oral feeding (postoperative)	16 (5–37) days
Anastomotic leakage (n,%)	5 (46%)
Esophago-bronchial fistula	1 (9%)
No. of dilatations (n, range)	6 (2-20)
Fundoplication (n,%)	10 (91%)
Age at fundoplication (median, range)	3.9 months (1.8–6.6)
Redo fundoplication (n,%)	6 (55%)
Weight-for-height z-score (SD)	-0.80 (-2.20–2.40)

Tracheomalacia

Preoperative bronchoscopy was not performed in the first TTT patient, because it was not yet introduced as routine care for EA patients in our clinic. Six from the subsequent 10 patients were diagnosed with tracheomalacia during their evaluation of the airway with preoperative bronchoscopy.

Two patients had severe postoperative tracheomalacia related symptoms, requiring aortopexy. One patient required thoracoscopic aortopexy at the age of 11 weeks and needed a redo thoracoscopic aortopexy at the age of 16 weeks. In the other patient a thoracoscopic aortopexy was performed at the age of 19 months.

Early gastrointestinal outcome

First oral feeding was started at a median of 16 days after performing the esophageal anastomosis (range 5-37). In one patient with Down syndrome with postoperative anastomotic leakage and respiratory instability, oral feeding was introduced after 37 days. All patients required multiple dilatations for anastomotic stenosis. The majority of dilatations (80%) was performed within the first year of life. A fundoplication was performed in 10 patients (91%) at a median age of 3.9 months (range 1.8-6.6 months). Six of these 10 patients required a redo fundoplication after a median of 6.3 months (range 2.9-58.7 months) (**Table 3**). The patient with Down syndrome later developed an esophagobronchial fistula, which was closed at the age of one year.

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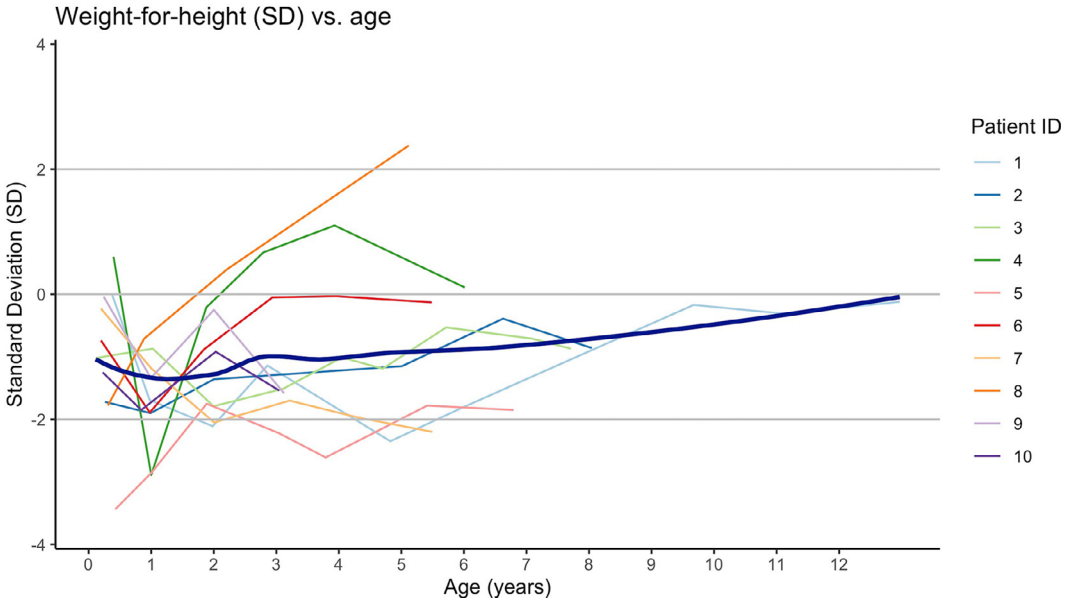

Fig. 1. Weight-for-height z-score of individual patients (n = 10). The dark blue line represents the mean weigh-for-height z-score of the whole group.

Follow-up

Reflux symptoms

The median age at follow-up was 7.0 years (range 3.3–13.4). Nine patients (82%) were using GER medication at time of this study. Nine out of ten subjects filled out the reflux-questionnaires (90%). Three patients (33%) reported no GER complaints, five patients (56%) reported mild complaints and one patient (11%) had moderate GER complaints. Of the patients that reported no reflux symptoms, all had a fundoplication and two a redo-fundoplication. Of the five patients that reported mild symptoms, four patients had a fundoplication of three of which also had a redo-fundoplication. The patient with moderate reflux symptoms had both a fundoplication and a redo-fundoplication.

Oral intake

Seven patients (70%) had no food limitations (FOIS 7), one patient had specific food limitations (difficulties with carrots and meat, FOIS 6) and one patient required special preparation of food (thickening of liquids, FOIS 5), but had no other food limitations. One patient with Down syndrome had achieved full oral intake, but had later regressed to tube-feeding with minimal attempts of oral food intake (FOIS 2).

Growth

Most patients had a decrease in weight-for-height z-score within the first year of life and a catch-up in the weight-for-height z-sore over time; the median weight-for-height z-score at the age of 1 year was -1.77 (range -2.89 to -0.71), compared to a median weight-for-height z-score at last follow-up of -0.80 (range -2.20 to 2.40) (n = 10). **Fig. 1** shows the weight-for-height z-scores over time for all included patients.

At end of follow-up, one patient had a weight-for-height z-score below -2SD due to unknown causes. Another patient had a weight-for-height z-score above 2SD, due to a small height and a normal weight-for-age. All other patients (80%) were within normal growth range.

12.1 Health-related quality of life

Nine out of ten PedsQL™ 4.0 questionnaires (90%) were returned. HRQoL parent-proxy report scores for patients younger than 5 years-old were higher than the cut-off points (1SD below the mean) on all domains (**Fig. 2a**).

The mean total score in patients older than 5 years was 80.1 for parent-proxy and 80.8 for child self-report (mean difference 0.7, 95%CI -5.2-3.7). These total scores were similar to the means of healthy controls (81.3 and 82.9, respectively). Both child self-report and parent-proxy report scored lowest on the school functioning domain (median of 69 and 68 respectively, compared to 77 and 80 in healthy controls), but above the threshold of 1 SD below the mean (67 and 62, respectively). Patients scored best on the social functioning domain (93 and 88, respectively) (**Fig. 2b**).

One patient scored below the PedsQL™ 4.0 total score cut-off point with a score of 63.0 on child-self report, which is 6.7 points below the score cut-off point of 69.7. His total lowered score was mainly due to a low score on the emotional and school functioning domain.

Discussion

This is the first study to evaluate the childhood outcome of LGEA patients treated with the thoracoscopic traction technique. TTT was successful in 11/13 patients (85%). This study shows that after TTT patients are able to initiate oral feeding as soon as 16 days after esophageal correction and almost all patients achieved an age-appropriate oral diet and growth patterns within normal range. The overall HRQoL is comparable to healthy children. This study further shows that reflux is common in LGEA patients after TTT. All patients required multiple dilatations for anastomotic stenosis and almost all patients (91%) required a fundoplication.

Postoperative stenosis is the most frequent postoperative complication after EA repair [12]. In this study, all LGEA patients needed multiple dilatations for recurrent anastomotic stenosis. A high stenosis rate in LGEA patients after TTT may be explained by the risk factors for anastomotic stenosis, including anastomotic tension, leakage and GER [24–27]. All patients needed multiple dilatations. Most dilatations (80%) were performed within the first year of life.

EA patients have a greater risk of developing GER and this is especially common in LGEA patients. It has been reported in 66-88% of LGEA patients after delayed primary anastomosis (DPA) [28,29].

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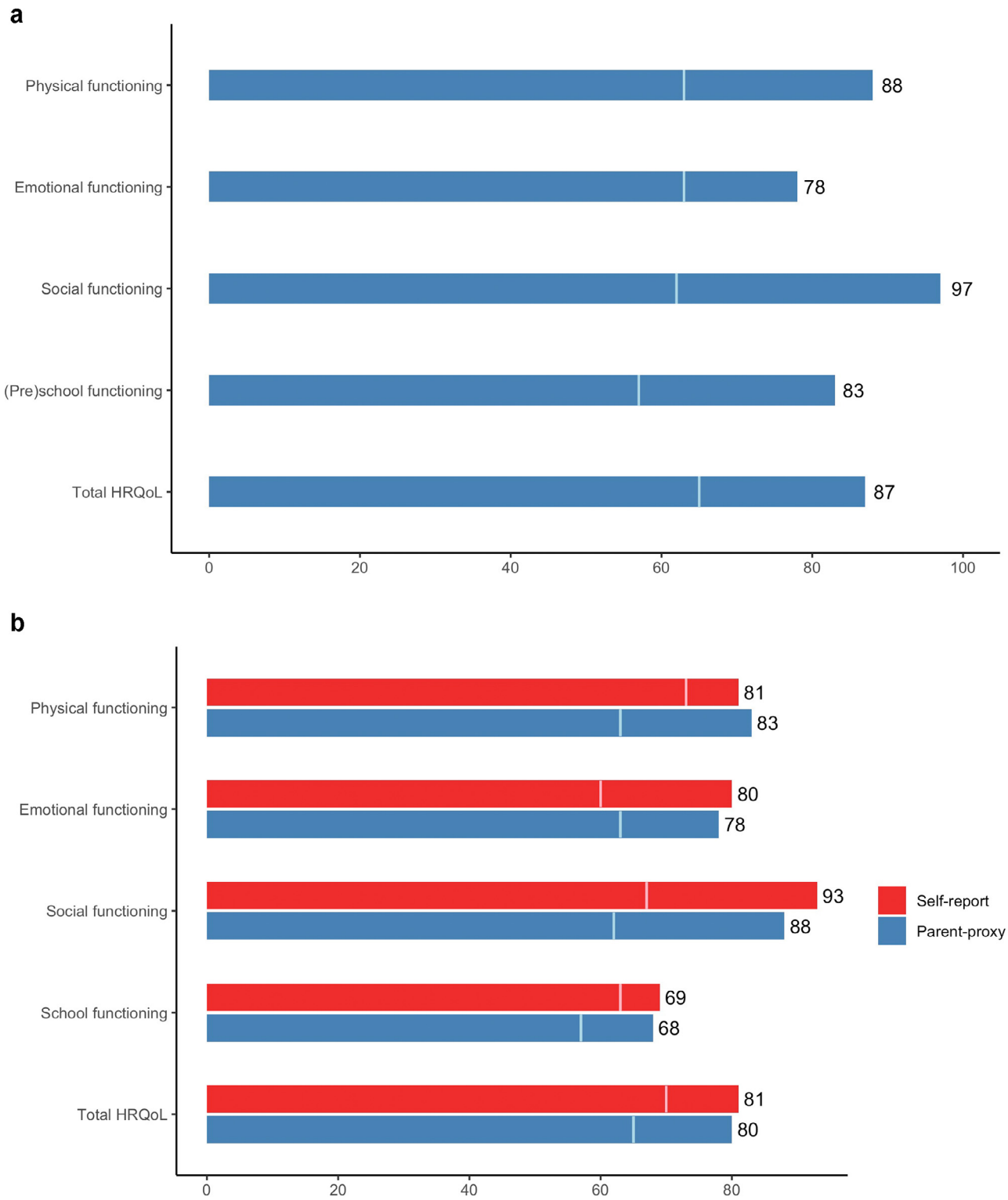

Fig. 2. a. HRQoL in patients <5 years old (parent-proxy report). b. HRQoL in patients ≥5 years-old (self-report and parent-proxy report). The vertical light lines represent scores of the cut-off point of 1 SD below the mean in the healthy population.

This is in line with our findings, in which GER symptoms were reported in 67%.

In this study, esophageal anastomosis was performed at a median age of 12 days. Oral feeding could be started 16 days postoperatively. In delayed primary anastomosis, the anastomosis is usually performed at the age of two to three months [12]. Therefore, oral feeding can only be introduced thereafter and patients will be fed by a gastrostomy in the period before esophageal anastomosis [12]. Feeding difficulties are common in EA patients and include eating slowly, food refusal and choking [30–32]. Since infants develop their feeding and swallowing skills within the first two years of life, later introduction of oral feeding may lead to delayed positive oral experiences [33,34]. Consequently, later introduction of an oral diet may impair the development of adequate feeding and

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swallowing skills [34,35]. Cavallaro et al. [15] reported severe feeding problems after DPA in five patients compared to no feeding problems in 20 EA-TEF patients. Bevilacqua et al. [33] showed that LGEA was associated with not reaching self-feeding at the age of 3 years. This is in contrast to our study, showing that almost all patients achieved an age-appropriate oral diet. Therefore, early introduction of oral feeding seems to be an advantage of TTT compared to DPA. We believe early oral feeding may contribute to patientsóral feeding performance and reduces the long term feeding difficulties that are common in LGEA patients.

Moreover, due to an early esophageal anastomosis (median day 12), patients do not require a preoperative gastrostomy, which may be associated with a high complication rate [36].

The total hospital length of stay is subsequently shorter in TTT compared to DPA (47 days vs. 120-150 days) [28,29].

Previous studies have reported that EA patients are at risk for growth problems, especially within the first years of life [22,37,38]. This is in line with our findings, which showed a decrease in weight-for-height z-scores within the first year of life. However, a catch-up in weight-for-height z-scores was seen over time. Almost all children were within normal growth range (-2SD and 2SD) at end of follow-up, although nutritional status in most children was still below the population mean.

In line with our findings, Peetsold et al. [39] reported a similar HRQoL in EA patients compared to healthy controls. Dingemann et al. [40] studied HRQoL in complex and complicated EA, including DPA, and showed a HRQoL comparable to healthy controls. Legrand et al. [41] reported that the QoL in EA patients is lower compared to healthy controls, but higher compared to patients with other chronic diseases. Our study shows that the overall HRQoL is comparable to healthy controls.

The main limitation of this study is the small sample size, which makes statistical comparison to healthy controls impossible. However, LGEA is a rare anomaly and only patients older than two years of age that had pure LGEA (Gross type A and B) were included. Ideally, a prospective multicenter study should be conducted to increase the sample size and to evaluate and compare the long-term outcome of the different techniques used for esophageal repair in LGEA. However, since the rarity of this disease, it might proof to be very difficult to conduct such a prospective study.

A second limitation of this study entails the wide study period, in which we changed from paper records into digital records, therefore some data could not be included (duration of parental nutrition, durations central venous catheter dependency).

In conclusion, TTT was successful in 13 of 15 patients (85%). Major advantages of the TTT are preservation of the native esophagus, early introduction of oral feeding and a shorter total hospital length of stay. Almost all patients are able to eat an age-appropriate oral diet and have a growth pattern within normal ranges. Feeding problems later in life may be prevented by TTT. Overall HRQoL in LGEA patients treated with TTT is comparable to healthy controls.

Declarations of Competing Interest

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