Pediatric Radiology (2022) 52:468–476
https://doi.org/10.1007/s00247-021-05228-z

ORIGINAL ARTICLE

Radiographic assessment of traction-induced esophageal growth and traction-related complications of the Foker process for treatment of long-gap esophageal atresia

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Received: 27 May 2021 / Revised: 19 September 2021 / Accepted: 12 October 2021 / Published online: 30 November 2021

Abstract

Background Radiographic assessment of esophageal growth in long-gap esophageal atresia while on traction and associated traction-related complications have not been described.

Objective To demonstrate how chest radiography can estimate esophageal position while on traction and to evaluate radiography's utility in diagnosing certain traction system complications.

Materials and methods In this retrospective evaluation of portable chest radiographs obtained in infants with long-gap esophageal atresia who underwent the Foker process between 2014 and 2020, we assessed distances between the opposing trailing clips (esophageal gap) and the leading and trailing clips for each esophageal segment on serial radiographs. Growth during traction was estimated using longitudinal random-effects regression analysis to account for multiple chest radiograph measurements from the same child.

Results Forty-three infants (25 male) had a median esophageal gap of 4.5 cm. Median traction time was 14 days. Median daily radiographic esophageal growth rate for both segments was 2.2 mm and median cumulative growth was 23.6 mm. Traction-related complications occurred in 13 (30%) children with median time of 8 days from traction initiation. Daily change >12% in leading-to trailing clip distance demonstrated 86% sensitivity and 92% specificity for indicating traction-related complications (area under the curve [AUC] 0.853). Cumulative change >30% in leading- to trailing-clip distance during traction was 85% sensitive and 85% specific for indicating traction complications (AUC 0.874).

Conclusion Portable chest radiograph measurements can serve as a quantitative surrogate for esophageal segment position in long-gap esophageal atresia. An increase of >12% between two sequential chest radiographs or >30% increase over the traction period in leading- to trailing-clip distance is highly associated with traction system complications.

Keywords Chest · Congenital · Esophageal atresia · Esophagus · Foker process · Infants · Radiography · Surgery

Introduction

Esophageal atresia is the most common congenital anomaly of the esophagus, occurring in about 1 in 4,100 live births [1]. Long-gap esophageal atresia has broadly been defined as a subset of esophageal atresia in which the distance between the proximal and distal esophageal segments is too far for primary surgical repair [2]. The Foker process for the surgical treatment of long-gap esophageal atresia utilizes continuous longitudinal traction that is intermittently increased to pull each esophageal segment to stimulate esophageal growth. At a cellular level, such strain on the esophagus increases nuclear density, cellular proliferation and myogenin expression while maintaining stable esophageal wall

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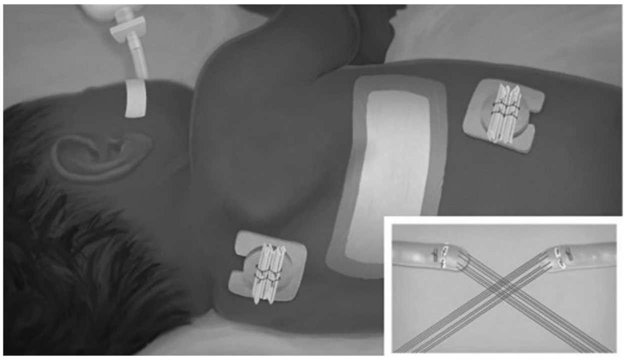

Fig. 1 Clinical image demonstrates the external portion of the Foker traction system with the traction sutures tied to small pieces of feeding tubes along the posterior and lateral chest to induce gentle longitudinal tension

thickness [3–5]. The Foker process is a technically demanding multi-step process with potential for significant complications including esophageal leak, traction system disruption or malfunction requiring reoperation [6, 7].

During the first stage of the Foker process (Foker I procedure), the proximal and distal esophageal segments are identified, either thoracoscopically or via open incision, and a series of four to six traction clips are placed on the ends of each esophageal segment. Pledget sutures connected to these traction clips are brought out of the infant's chest and tied onto an external traction system (Fig. 1). These traction sutures are tightened every 2–3 days at the bedside to generate longitudinal traction on both the proximal and distal esophageal segments. Portable chest radiographs are acquired immediately following the Foker I procedure and routinely after suture tightening, usually every 1–3 days [8]. When there is sufficient overlap of the esophageal segments, esophageal continuity is established by performing an esophageal anastomosis (Foker II procedure).

To monitor esophageal growth and detect potential traction system complications, each esophageal segment is marked by two types of radiopaque metallic clips, which can be sequentially followed with portable chest radiographs. Small radiopaque clips (leading clips) mark each traction suture at the esophageal segment tip (Fig. 2). A single large metallic clip (trailing clip) placed slightly proximal to the traction sutures denotes the position of the esophagus and is not subject to direct traction (Fig. 2). As the traction sutures are tightened externally, leading and trailing clips of each segment should move in tandem on subsequent radiographs (Fig. 2).

Radiographic evaluation of the Foker process has been described; however, details of the radiographic assessment of the Foker traction system, the growth process and the specific traction complications have not been described [9]. The purpose of this study was to demonstrate a radiographic method to estimate esophageal growth for children with long-gap esophageal atresia treated with the Foker process, and to evaluate its utility in diagnosing the imaging appearance of certain traction system complications.

Materials and methods

Our institutional review board approved this retrospective review of surgical and imaging data of infants with long-gap esophageal atresia treated at our institution from 2014 to 2020. Informed consent was waived because the retrospective data included in this study were acquired as part

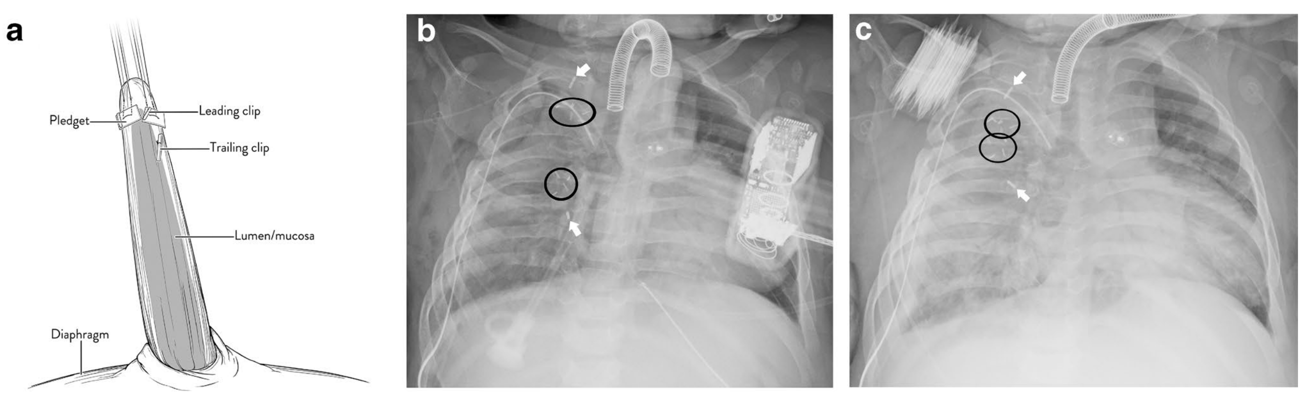

Fig. 2 Esophageal leading and trailing clips placement. **a** Schematic drawing shows the position of the leading and trailing clips on the distal esophageal segment. Suture threads attached to the leading clips and pledget (not radiographically visible) are used to apply longitudinal tension on the esophageal segment. Radiographically, the leading clips are a group of small clips at the tip of each esophageal segment (usually 4–6 clips) and the lagging clip is the single larger clip proximal to the leading clips of each segment. **b, c** Portable anteroposterior chest radiographs in a 16-month-old girl with VACTERL (vertebral, anorectal, cardiac, trachea-esophageal, renal, limb anomalies) association at postoperative day 0 (**b**) and postoperative day 5 (**c**) demonstrate the appropriately positioned leading (*circles*) and trailing (*arrows*) clips moving in tandem

of routine clinical management and the risk to the patients was minimal.

Study cohort

We evaluated for inclusion all children with long-gap esophageal atresia who underwent repair at our institution between 2014 and 2020. We excluded children who underwent long-gap esophageal atresia repair by means other than the Foker process using external traction or who had initial postoperative chest radiographs with overlap of the proximal and distal segment suture markers preventing clear segment identification (Fig. 3). Demographic information included age, gender, weight, esophageal atresia type, presence or absence of VACTERL (vertebral, anorectal, cardiac, trachea-esophageal, renal, limb anomalies) association, time on traction, and development of a traction complication. Clinical data and surgical outcomes for the cohort included in this study and for additional children with long-gap esophageal atresia treated with alternative surgical techniques have recently been published; however, none of the radiographic assessment data have been described [8].

Image acquisition and radiographic measurements

All portable chest radiographs were acquired with a source-to-detector distance of 40 in., a tube voltage of 60–68 kVp (depending on subject size) and a tube current of 0.8 mAs.

To achieve overlap during the Foker process, the esophageal segments are pulled in a criss-cross orientation rather than end-to-end. This results in variable and inaccurate gap measurements if one were to measure in a direct line from trailing clip to trailing clip. As such, a baseline parallel to the closest adjacent vertebral body inferior endplate was placed along the inferior margin of the lower esophageal segment trailing clip to allow for more consistent measurements (Fig. 4). By doing so, the distance between the esophageal segments, or esophageal gap, was measured as a line drawn perpendicular from the baseline to the most superior margin of the opposing trailing clip (electronic calipers, Fuji Picture Archiving and Communication System [PACS],

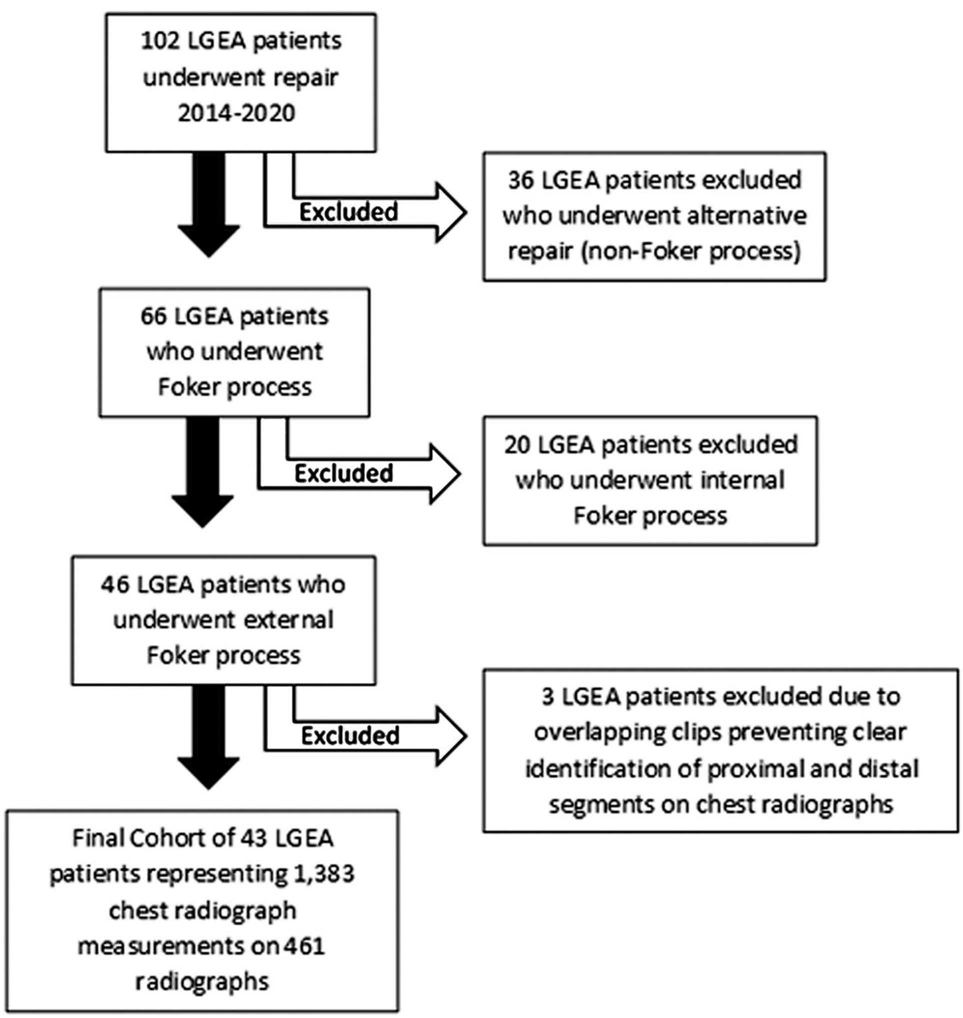

Fig. 3 Flow diagram shows selection of the long-gap esophageal atresia (*LGEA*) cohort

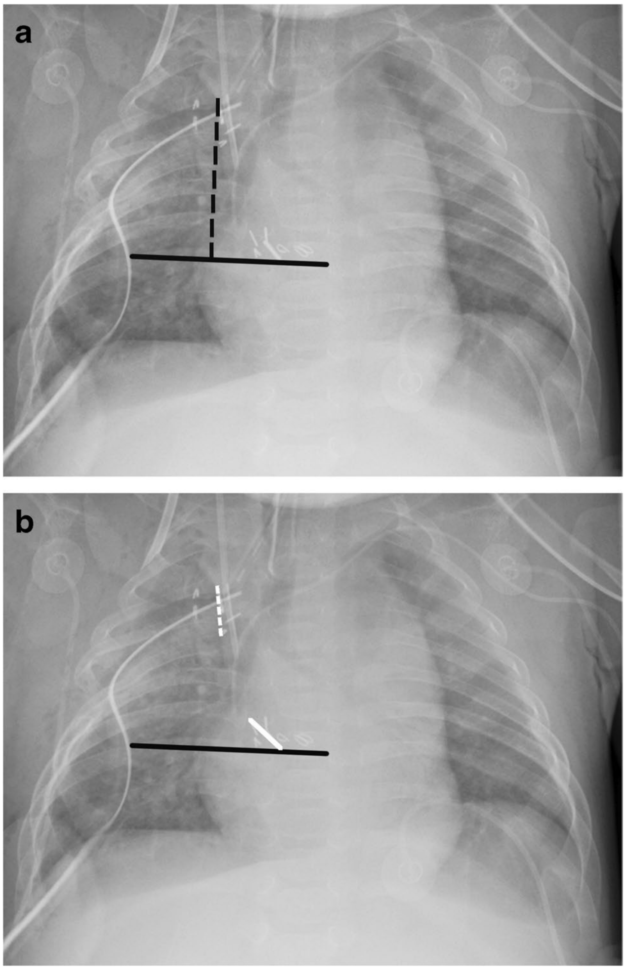

Fig. 4 A single portable anteroposterior chest radiograph of a 4-month-old boy with long-gap esophageal atresia undergoing an external Foker process is shown twice to demonstrate measurement technique. **a** baseline (*solid black line*) is drawn at the inferior margin of the lower segment trailing clip parallel to the closest adjacent vertebral body inferior endplate to allow for a consistent starting point for the measurements. Subsequently, the distance from the baseline (*solid black line*) to the superior margin of the upper segment trailing clip is measured (esophageal gap; *black dashed line*). **b** The leading- to trailer-clip distance for each segment is then measured as the greatest distance between the distal-most part of the trailing clip and the proximal end of the most proximal leading clip regardless of clip orientation (*solid and dashed white lines*)

Fujifilm North America Corp., Valhalla, NY) (Fig. 4). We measured the distance between the most peripheral aspect of the distal leading traction clip and the most peripheral aspect of its corresponding trailing clip (leading- to trailing-clip distance) for each esophageal segment to assess traction system/esophageal integrity (electronic calipers, Fuji PACS) (Fig. 4). As the esophageal segments grow, the distance between the leading clips and marker clip should remain relatively constant in an intact system, thus loss of this relationship suggests a possible traction complication.

Electronic radiographic measurements were recorded by a single board-certified pediatric trained radiologist (1 year post fellowship, A.M.F.) for each chest radiograph obtained between Foker I and Foker II stages for each child to assess the esophageal gap or distance between the trailing clips, as well as the relationship between the leading and trailing clips of each esophageal segment (Fig. 4). A random sample of measurements were repeated independently in a blinded fashion by the same radiologist and also by a second board-certified pediatric radiologist (20 years post fellowship, M.J.C.) to assess intra- and inter-rater reliability.

Traction-related complication definitions

We defined traction-related complications as follows: (1) In suture dislodgement, one or more isolated traction sutures breaks or tears directly from the esophageal tip, reflected by one or more leading clips significantly separating from the rest (Fig. 5). (2) In cap rupture, the entire tip of the esophagus sustains a muscular disruption and all the leading clips travel together and separate abruptly and drastically from the trailing clip (Fig. 6); pneumomediastinum or worsening pleural effusions might be observed. (3) Mucosal tube is when the muscular esophageal wall is disrupted, but with intact mucosa connected to the tip of the esophagus that continues to grow along with the tip, demonstrating a more gradual separation of the leading and trailing clips (Fig. 7). (4) Leak on traction occurs when mucosa is breached and either saliva (if upper segment) or gastric/intestinal secretions (if lower segment) leak into the thoracic cavity. Despite traction sutures being carefully placed with endoscopic guidance, a leak on traction can develop with any of these scenarios.

Statistical analysis

From the measurements for leading- to trailing-clip distance of each esophageal segment, we calculated a daily percentage change and a cumulative percentage change (the sum of each daily percentage change over the course of the traction period). We then assessed differences in daily and cumulative percentage change among infants with and without a traction system complication, and among the different types of traction system complications using the Mann–Whitney *U* test. We calculated the median daily esophageal growth on traction both empirically and also using a generalized least squares regression model accounting for serial radiograph measurements within the same child [10]. A random-effect was implemented to account for multiple and varying numbers of radiographs per child to control for variability among children and within–patient correlation. To establish growth thresholds that could be associated with development of

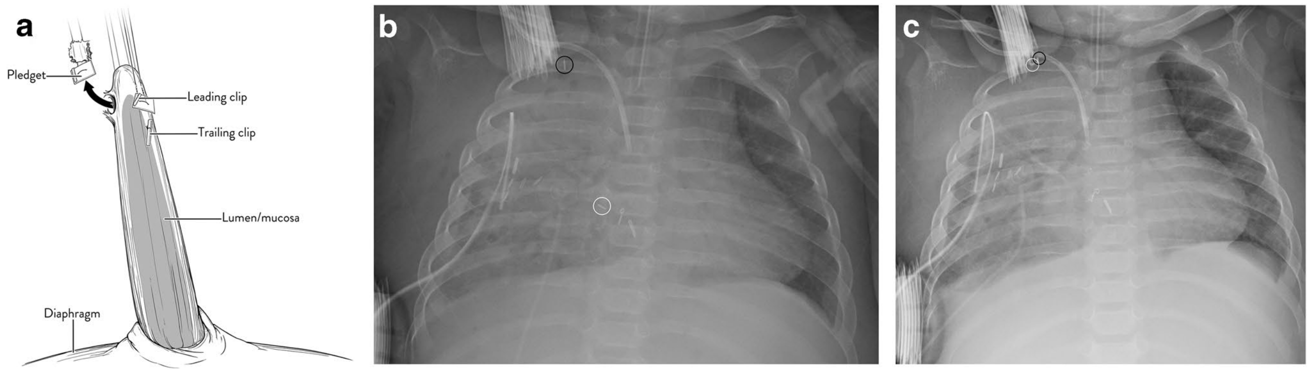

Fig. 5 Suture dislodgment. **a–c** Schematic diagram (**a**) and portable anteroposterior chest radiographs in a 6-month-old girl (**b, c**) show suture dislodgement. Postoperative day 0 image (**b**) shows that one leading clip from the lower segment has become dislodged (*black circle*); note normally positioned clip (*white circle*). On postoperative day 2 in the same girl (**c**), a second lower-segment leading clip has also become dislodged (*white circle*)

traction system complication, we performed a multivariable logistic regression and constructed receiver operating characteristics (ROC) curves with the Youden J index to identify optimal cut-off values [11]. We calculated intraobserver and interobserver agreement using the intraclass correlation coefficient (ICC) based on a two-way mixed-effects model of absolute agreement with a random sample of 20 radiographs (2 random radiographs from 10 random patients) [12]. We selected the sample of 10 random patients based on a uniform (0,1) distribution by implementing the "runiform" function in Stata software (version 16.0; StataCorp, College Station, TX) and giving each of the 43 children an equal probability of random selection. Then, radiographs were randomly selected from each of these 10 children, again based on a uniform (0,1) distribution.

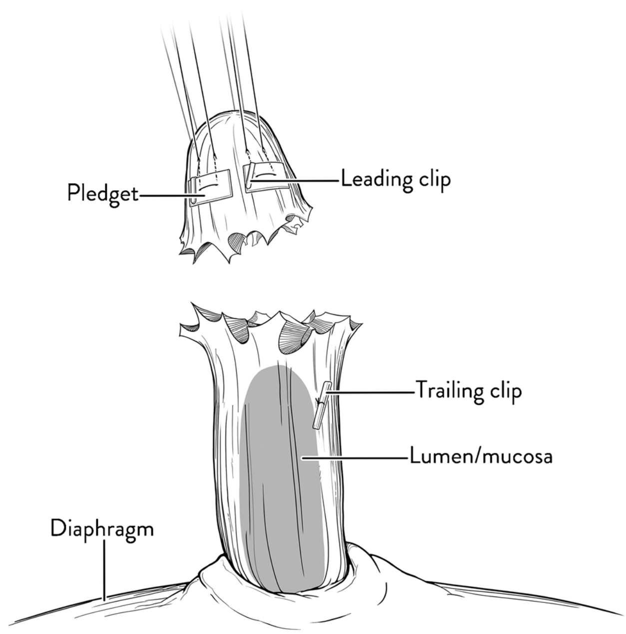

Fig. 6 Schematic diagram shows an esophageal segment cap rupture

Two-tailed values of $P<0.05$ were considered statistically significant. A sample size of 461 chest radiographs from 43 children provided 80% power for estimating daily esophageal growth on traction to within 0.3 mm per day based on a generalized least squares regression model, assuming a two-tailed 5% alpha.

Results

From 2014 to 2020, 43 children (25/43 [58%] male) who underwent the Foker process via external traction at our institution for long-gap esophageal atresia met inclusion criteria for this study. Demographic data are presented in Table 1. The median esophageal starting gap distance was 4.5 cm (interquartile range [IQR] 3.7–5.5 cm).

The main outcome measures are shown in Table 2. Median time on external traction was 14 days (IQR 10–17 days) and the average number of days between traction adjustments was 2.3 days (IQR 1.8–2.8 days, range 1.3–5 days). A complication with the traction system occurred in 13 (30%) of the children, including suture pull (*n*=8, 18.6%), traction leak (*n*=3, 7.0%) and mucosal tube (*n*=2, 4.6%). Such traction-related complications occurred at a median of 8 days (IQR 4–12 days) from the time of the Foker I or traction system setup. The clinical outcomes of these children have been described [8].

Radiographic assessment during the Foker process

We reviewed a total of 461 chest radiographs (average 11, range 2–28 per child), which were performed while infants were on external traction. Portable chest radiographs were

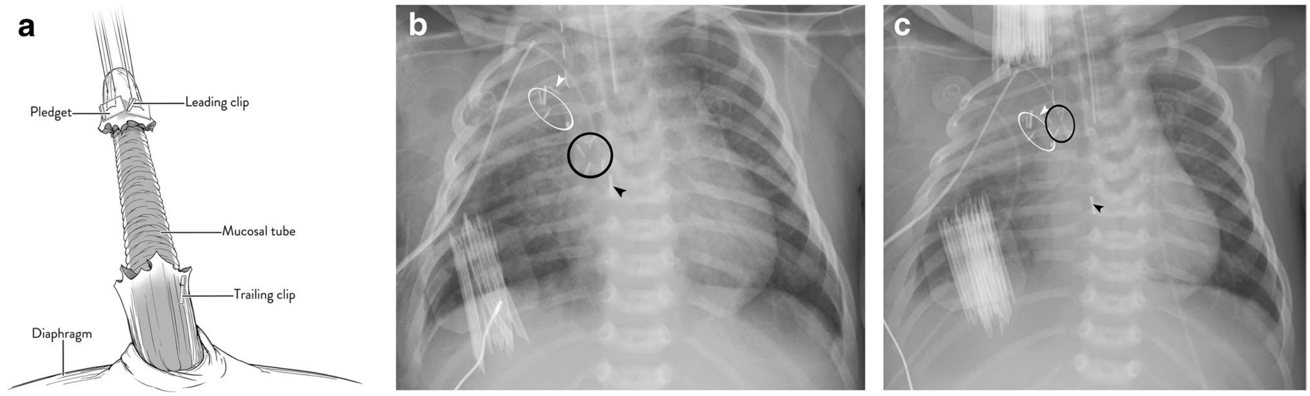

Fig. 7 Mucosal tube. **a–c** Schematic diagram (**a**) and portable anteroposterior (AP) chest radiographs in a 26-day-old girl (**b, c**) demonstrate formation of a mucosal tube. AP chest radiograph on postoperative day 0 (**b**) shows the leading clips (*ellipses*) and trailer clips (*arrowheads*) appropriately positioned within the upper (*white annotations*) and lower (*black annotations*) esophageal segments. On postoperative day 2, AP chest radiograph (**c**) shows the leading- to trailer-clip distance for the lower segment pouch has substantially increased without pneumomediastinum because of a mucosal tube (confirmed intraoperatively). Segmentation anomalies in the vertebrae of the mid thoracic spine are also evident

performed primarily to evaluate a bedside traction system adjustment (272/461, 59%), after an operation or reoperation to establish a traction system baseline (60/461, 13%), or for a non-traction-related clinical indication (129/461, 28%) such as an endotracheal tube adjustment, patient desaturation, ventilator adjustment or to monitor fluid status. Intraobserver and interobserver consistency for each of the three radiographic measurements were excellent, with ICC values greater than 0.97 for all radiographic measurements (all $P<0.001$; Table 3).

Radiographic assessment of esophageal growth

Median daily rate of esophageal growth (measured radiographically as the shortening of the inter-trailer clip distance) was 2.2 mm (IQR 0.8–4.2 mm) for both segments, or

Table 1 Demographics of the long-gap esophageal atresia study cohort

Demographics	
Number of patients	*n*=43
Age at surgery (months)	Median: 5, IQR: 3–6, range: 0–22
Male/female gender	*n*=25/18
Weight (kg)	Median: 5.8, IQR: 4.0–6.8, range: 3.3–10.9
Prematurity	
30–37 weeks of gestation	*n*=25 (58%)
<30 weeks of gestation	*n*=4 (9%)
Term	*n*=14 (33%)
Esophageal atresia gross type	
A (with no fistula)	19 (44%)
B (with proximal fistula)	9 (21%)
C (with distal fistula)	14 (33%)
D (with proximal and distal fistula)	1 (2%)
E (no atresia with fistula)	0 (0%)
Isolated esophageal atresia	10 (23%)
Esophageal atresia with VACTERL[a]	33 (77%)
Starting esophageal gap (mm)	Median: 45, IQR: 37–55, range: 20–70
Rescue Foker	12 (28%)

IQR interquartile range, *VACTERL* vertebral, anorectal, cardiac, trachea-esophageal, renal, limb anomalies

[a]Two or more anomalies need to be present to qualify for VACTERL association

Table 2 Main outcome measures

Results	
Time on external traction (days)	Median: 14, IQR: 10–17
Days between traction adjustments	Median: 2.3, IQR: 1.8–2.8
Daily esophageal growth (mm)	Median: 2.2, IQR: 0.8–4.2, average: 1.8 mm
Percentage daily growth (%)	
Suture pull	Median: 27, IQR: 12.3–38.5
Mucosal tube	Median: 62, IQR: 57–88.7
Traction leak	Median: 49.6, IQR: −13.4–80.2
No traction complication	Median: 0.5, IQR: −1.3–4.8
Cumulative esophageal growth (cm)	Median: 23.6, IQR: 14.8–32.0
Percentage cumulative growth (%)	
Suture pull	Median: 53.9, IQR: 35.5–85.7
Mucosal tube	Median: 82.6, IQR: 71.7 to 99.2
Traction leak	Median: 18.5, IQR: −5.9–30.9
No traction complication	Median: 14.1, IQR: −3.7–22.5
Traction problem	13 (30.2%)
Suture pull	8 (18.6%)
Traction leak	3 (7%)
Mucosal tube	2 (4.6%)

IQR interquartile range

1.1 mm per esophageal segment. Longitudinal analysis using a random-effects generalized least squares regression model to account for serial radiographic measurements within the same child indicated an average esophageal growth rate of 1.8 mm per day on traction (P<0.001). Median cumulative growth for the entire traction period was 23.6 mm (IQR 14.8–32.0 mm), or 11.8 mm per esophageal segment.

Radiographic assessment of traction system complications

The median (IQR) daily percentage change of the leading- to trailing-clip distance per esophageal segment was significantly greater in infants with a traction system complication. Specifically, median daily percentage change was greater for infants with a suture pull (27% change, IQR 12–39%) or a mucosal tube (62% change, IQR 57–89%) compared to those without a traction complication (0.5% change, IQR −1% to 5%, P<0.01). However, there was no significant difference between those with a traction leak (50% change, range −13% to 80%) and the group without a traction complication (P=0.29). The optimal cutoff point for daily percentage change of >12% resulted in a sensitivity of 86% (12/14) and a specificity of 92% (839/908) with an area under the ROC curve of 0.853 (95% confidence interval [CI]: 0.703–0.999) (Fig. 8).

Similarly, when evaluating the entire traction time period with the cumulative percentage change of the leading- to trailing-clip distance per esophageal segment, infants with a traction system complication had greater (median, IQR)

Table 3 Intraobserver and interobserver agreement

Measurement	ICC	95% CI	P-value	SD of difference	2 SD of difference (95% CI)
Intraobserver agreement (within the same observer)					
Distance between	0.998	(0.994, 0.999)	<.0001	1 mm	2 mm
Distance proximal pouch	0.975	(0.938, 0.989)	<.0001	0.7 mm	1.4 mm
Distance distal pouch	0.994	(0.984, 0.998)	<.0001	0.6 mm	1.2 mm
Interobserver agreement (between observers)					
Distance between	0.990	(0.975, 0.996)	<.0001	2 mm	4 mm
Distance proximal pouch	0.978	(0.945, 0.991)	<.0001	0.7 mm	1.4 mm
Distance distal pouch	0.992	(0.980, 0.997)	<.0001	0.7 mm	1.4 mm

CI confidence interval, *ICC* intraclass correlation coefficient, *SD* standard deviation

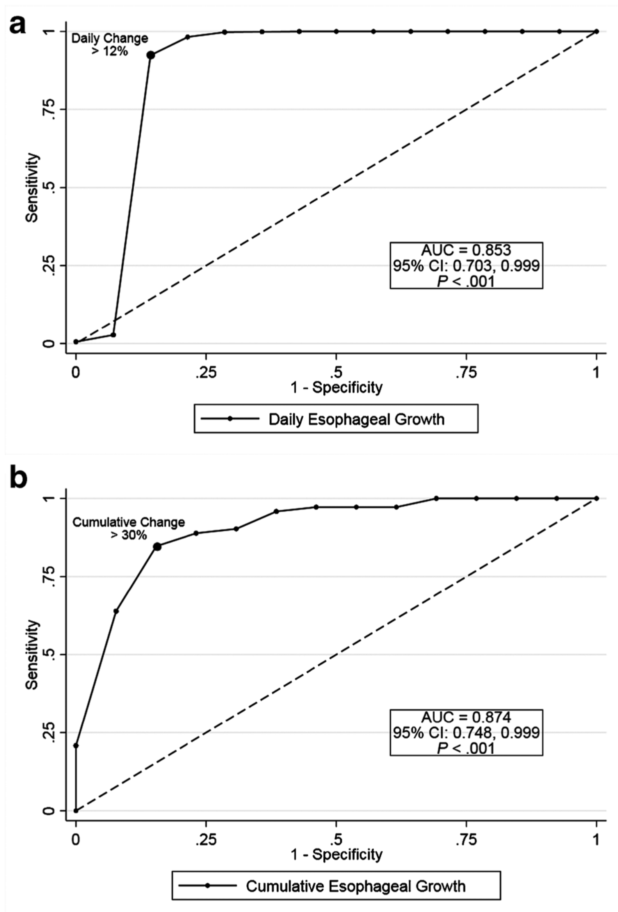

Fig. 8 Receiver operating characteristics curves for traction-related complications. **a, b** Daily change in leading- to trailer-clip distance >12% provides 86% sensitivity and 92% specificity for determining traction complications (**a**), and cumulative change in leading- to trailer-clip distance >30% demonstrates 85% sensitivity and 85% specificity for determining traction complications (**b**). Daily and cumulative growth were strong discriminators for classifying cases with and without traction complications. *AUC* area under the curve, *CI* confidence interval

cumulative percentage changes, specifically when comparing those with a suture pull (54%, 36–86%) or mucosal tube (83%, 72–99%) with those without a traction complication (14%, −4% to 23%) ($P<0.01$ for both). Again, those with a leak on traction (19%, −6% to 31%) had similar cumulative percentage change values to those without a traction complication ($P=0.73$). The optimal cutoff point for cumulative percentage change of >30% resulted in a sensitivity of 85% (11/13) and specificity of 85% (61/72) with an area under the ROC curve of 0.874 (95% CI: 0.748–0.999) (Fig. 8).

Discussion

Long-gap esophageal atresia represents one of the most complex and challenging clinical conditions faced in pediatric surgery. The concept that "one's own native esophagus is best" was the cornerstone for the evolution of tension-accelerated natural growth first described by Dr. John Foker in 1997 [6]. Since that time, the Foker process has evolved, with continued improvement in patient outcomes [8]. As more centers begin to perform this technique, it is important to describe the known traction-related complications of the external Foker process [7, 13]. This study illustrates the use of diagnostic medical imaging in estimating esophageal growth and demonstrates the radiographic manifestations of traction system complications.

Esophageal segment growth has been demonstrated clinically; however, this study provides a quantitative surrogate measurement for growth rate of the esophageal segments using medical imaging. Establishing an approximate growth rate is important because it can help surgeons more accurately estimate the time on traction that might be required for a particular case based on the starting gap length.

The surgeon performing the tension adjustment and radiologist evaluating the postoperative radiographs must maintain a high index of suspicion for complications. To this end, this study highlights the importance of closely evaluating the position of the leading and trailing clips relative to each other for both esophageal segments across radiographs obtained during the traction process. Specifically, the observation of a sudden or gradually progressive increase in the leading- to trailing-clip distance of greater than 12% between consecutive radiographs or of more than 30% during the traction process is highly sensitive and specific for a traction system complication. Although measuring an exact percentage of change might not be practical in day-to-day clinical work, careful scrutiny must be paid to leading and trailing clip positions compared to multiple recent prior examinations, with particular attention to changes in clip position. In the case of traction suture pull, the distance between one or more, but not all, of the leading clips and the trailing clip substantially increases. For a mucosal tube, the distance between of all of the leading clips and the trailing clip increases. Theoretically, a sudden increase between the group of leading clips and the trailing clip would also be observed with a cap rupture, although there were no cap ruptures in this study. Clinically, a cap rupture can be detected by noticing lack of tension on all of the traction sutures. Although there was no significant difference in leading- to trailing-clip distance between cases of traction-associated leak and cases without a traction-related complication, other clinical (fever, tachycardia, hypoxemia) and radiographic features such as extraluminal air, new or increasing pleural effusion or hydropneumothorax, or new cavitary lesion (developing abscess) might help identify traction-related leaks [9]. When in doubt, an esophagram while on traction can be performed to evaluate for leak.

This study has some limitations. A somewhat small sample size and a retrospective study design might limit generalizability of our results; however, given the rarity of long-gap esophageal atresia, this study is the largest such group evaluated radiographically. Second, differences in patient positioning might have introduced errors in measurement; however, given that the patients were similar in size and sedated and that images were acquired with a standard technique, differences should be minimal.

Conclusion

This study demonstrates that portable chest radiograph measurements can serve as a surrogate to estimate esophageal segment position in infants with long-gap esophageal atresia who are undergoing the Foker process. Furthermore, it reveals that an increase in leading- to trailing-clip distance of >12% between two sequential chest radiographs or >30% over the traction period is highly associated with traction complications. Complications require prompt clinical intervention, so it is important for interpreting radiologists to be aware of the relationship among these surgical clips across sequential chest radiographs.

Acknowledgments Dr. Alexandra M. Foust and Dr. Benjamin Zendejas contributed equally to this manuscript. Dr. Thomas E. Hamilton and Dr. Michael J. Callahan contributed equally as senior authors to this manuscript.

Declarations

Conflicts of interest None

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