



The burden of esophageal dilatations following repair of esophageal atresia



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ABSTRACT

Aim: To describe the burden of esophageal dilatations in patients following esophageal atresia (EA) repair.

Method: A retrospective review was performed at The Royal Children's Hospital, Melbourne, of all neonates undergoing operative repair for EA over a 17-year period (1999–2015). Stricture was defined by radiological and/or intra-operative findings of narrowing at the esophageal anastomosis. Data recorded included EA type, perinatal details, operative approach, esophageal anastomosis outcome, dilatation requirement, and survival. Key endpoints were anastomotic leakage and tension, esophageal dilatation technique, dilatation frequency, fundoplication, and complications.

Results: During the study period, 287 newborn EA patients were admitted, of which 258 underwent operative repair and survived to primary discharge. Excluding 11 patients with isolated tracheoesophageal fistula, 247 patients were included in the final analysis. Intra-operative anastomotic tension was documented in 41/247 (16.6%), anastomotic leak occurred in 48/247 (19.4%), and fundoplication was performed in 37/247 (15.0%). Dilatations were performed in 149/247 (60.3%). Techniques included bougie-alone (92/149, 61.7%), combination of bougie and balloon (51/149, 34.2%), and balloon-alone (6/149, 4.0%). These patients underwent 1128 dilatations; median number of dilatations per patient was 4 (interquartile range 2–8). Long-gap EA and anastomotic tension were risk factors ($p < 0.01$) for multiple dilatations. Complications occurred in 13/1128 (1.2%) dilatation episodes: 11/13 esophageal perforation, 2/13 clinically significant aspiration. Perforations were rare events in both balloon (6/287, 2.1%) and bougie dilatations (4/841, 0.5%); one patient had a perforation from guidewire insertion.

Conclusions: Esophageal dilatation occurred in a majority of EA patients. Long-gap EA was associated with an increased burden of esophageal dilatation. Perforations were rare events in balloon and bougie dilatations.

Type of study: Original article – retrospective review.

Level of evidence: II

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The burden of dilatation following esophageal atresia (EA) repair contributes to patient morbidity, due to stricture and dysmotility. The formation of an anastomotic stricture affects 18–50% of EA patients [1–7]. This may occur as early as 30 days post-operatively, or present many years following repair [3]. The pathogenesis of stricture formation

is complex and multifactorial. Suggested risk factors include long esophageal gap length, anastomosis under tension, two-layer anastomosis, anastomotic leak, eosinophilic esophagitis, and gastro-esophageal reflux disease (GERD) [1, 3, 5, 8–12]. Many factors may co-exist in the same patient, with some identifiable at the initial operation (gap length, anastomotic tension), whilst others occur post-operatively.

Esophageal atresia is also associated with foregut dysmotility, relating to abnormal development of the innervation and musculature of the esophagus, and to the esophageal repair [1, 13]. This plays a role in other comorbidities, including aspiration and respiratory complications, GERD, feeding issues, and dysphagia [14]. Up to 75% of EA patients have GERD

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[15]. Due to the complex interplay between these factors, symptoms may not resolve until all possible pathogenic mechanisms are addressed.

Over half of EA patients undergo intervention for anastomotic strictures or dysmotility – the majority with dilatation, and rarely, resection of stricture [16, 17]. There is currently no widely validated prognostic tool for risk stratification of patients post-repair [6].

Our study aimed to describe the burden of esophageal dilatations in EA patients following operative repair, and the associated risk factors. In addition, we aimed to consider what additional burden arises in this patient group due to complications of dilatation.

1. Methods

A single-center retrospective review of neonates undergoing operative repair of EA, during a 17-year period (January 1999 – December 2015) at The Royal Children's Hospital, Melbourne was performed. Surgery for EA repair was performed by nine surgeons at The Royal Children's Hospital during the study period. Each surgeon operated on three to four EA patients per year. All surgeons are consultant surgeons with a Fellowship in Pediatric Surgery from the Royal Australasian College of Surgeons. Trainees participated in operative repair, either as the primary operator or assistant, depending upon their level of experience. Throughout the period of the study, the surgical approach has been relatively static – this includes the selective use of chest drains and trans-anastomotic tubes. No minimally invasive repairs were performed at our institution during the study period. At the same time, there has been progression in the adjunctive neonatal care of EA patients. The management of EA patients at our institution has been well-documented in the literature [18, 19]. We utilized the prospectively-maintained Nate Myers Oesophageal Atresia database, with patient data from 1948 onwards. This database is maintained by the EA research nursing team, who are contacted during each hospital admission of EA patients, to maintain the database contemporaneously.

Data recorded included EA type (according to Gross classification), perinatal details, VACTERL (vertebral defects, anal atresia, cardiac defects, tracheoesophageal fistula, renal anomalies, limb abnormalities) association, operative approach, esophageal anastomosis outcome, survival, anastomotic tension and leakage, esophageal dilatation technique and frequency, and utilization of fundoplication. Patients with isolated tracheoesophageal fistula (Gross type E) were excluded, as they had no esophageal anastomosis, no tension, and therefore should not develop strictures. Anastomotic tension was reported by the operating surgeon. Any missing data were reported as such and excluded from further analysis.

Anastomotic leakage was defined as a pleural effusion or pneumothorax requiring intercostal drainage, ongoing output from intercostal drain placed at the initial thoracotomy, leak demonstrated during contrast study, and/or a clinical leak as documented in the patient's discharge summary [4]. Stricture was defined as those patients with subsequent radiological and/or intra-operative findings of narrowing at the esophageal anastomosis [3]. Tracheomalacia was defined by clinical diagnosis (eg, symptoms of stridor) or findings during endoscopic airway assessment (eg, flaccid tracheal cartilage).

Symptoms indicative of anastomotic stricture or dysmotility included swallowing difficulties (dysphagia, vomiting, cough, poor or slow feeding, acute life threatening events) and food bolus obstruction. All patients who presented with suggestive symptoms underwent rigid esophagoscopy to assess for stricture formation. Contrast studies were done at the discretion of the consultant surgeon. No routine prophylactic dilatations were performed. Diagnoses of GERD were based on symptomology, pH studies and/or esophageal biopsies. Routine use of Rantidine +/- proton pump inhibitors was initiated in the last decade.

Surgeon preference dictated which dilatation technique was used. All dilatations were undertaken under general anesthesia in the operating theater, and performed by a surgeon or interventional radiologist. The rigid dilatation technique involved the use of a 2.7 mm or 4.0 mm rigid esophagoscope to visualize the esophageal stricture. Pillings or

Savary-Gillard dilators (15–46 Fr) were used to dilate the esophagus. A guidewire and image intensification were used with Savary-Gillard dilators. In the narrowest strictures, some surgeons opted to re-purpose a Cook Medical ureteral dilator set with guidewire, as small as 8 Fr. A routine post-operative chest x-ray was not performed, though some patients had image intensification at the end of the procedure to check for complications.

Balloon dilatation involved the passage of a flexible guidewire through the esophageal stricture under fluoroscopic or endoscopic vision. The size of the dilator was determined with reference to the diameter of the esophageal stricture, character of the stricture, response to previous dilatation episodes and size of the child. The balloon dilator was inflated until the required pressure was achieved, and then held in the dilated position for approximately 3 min. The pressure of the balloon was then released and the procedure was repeated once again. Esophageal perforation was excluded by the instillation of contrast post-dilatation.

The frequency of, and interval between, dilatation sessions depended upon the evolution of symptoms suggestive of esophageal stricture or dysmotility. For the majority of patients, the technique was performed as a day case procedure. Patients were allowed to feed normally post-procedure.

The Nate Myers Oesophageal Atresia database, as well as this study's database, was maintained primarily by two authors (AH and JB in the former; JC and STT in the latter). This allowed for cross-checking to minimize the risk of data extraction errors. Statistical analysis was performed using SSPS IBM Corp (IBM SPSS Statistics for Windows, Version 22.0. Armonk, NY) and GraphPad Prism 7.00 for Windows (GraphPad Software, La Jolla, CA, USA). Categorical values were described with simple frequencies. chi-Square, *t*-test, and odds ratio calculation were used to test for significance and risks. Numerical variables were tested for normal distribution with the Shapiro-Wilk test, and described as median with range. Multinomial logistic regression was performed to assess for risk factors for dilatation. Approval from the Human Research Ethics Committee (RCH clinical audit approval QA/60161/RCHM-2019 and RCH project approval 35,089) was granted.

2. Results

A total of 287 patients were identified. From this initial group, 267/287 (93.0%) patients had an operative intervention and survived to discharge. Complete data, suitable for analysis, were available for 258/267 (96.6%) patients. There were 11 patients with isolated tracheoesophageal fistula, who were excluded – all of whom did not undergo esophageal dilatation. Therefore, a total of 247 patients were included in the final analysis.

The majority of patients were female (158/247, 64.0%), the median gestational age was 38 weeks (range 28–42 weeks), and the median birth weight was 2672 g (range 765 – 4450 g). There were no significant differences in these parameters across the different EA types (Table 1). Fifteen of 247 (6.1%) patients had a birth weight lower than 1500 g. A VACTERL association was identified in 58/247 (23.5%) patients. Tracheomalacia was documented in 52/247 (21.1%) patients. The number of patients with documented anti-acid medication on discharge was 187/247 (75.7%): 125/187 (66.8%) on proton pump inhibitors (PPIs) and 75/187 (40.1%) on H2-antagonists; 13/187 (7.0%) were discharged on both.

2.1. EA type

The predominant EA type was Gross type C (220/247, 89.1%), while Gross type A represented 14/247 (5.7%) of patients.

2.2. Type of repair

In 216/247 (87.4%) of patients, a primary repair was done, whilst 24/247 (9.7%) patients underwent a fistula division and delayed esophageal

Table 1
Patient demographics.

EA type	N (%)	Gestation (weeks)	Birth weight (g)	Primary repair (%)	VACTERL (%)
A	14 (5.4)	36 (34–38)	2383 (1780–2892)	-	1/14 (7.1%)
B	7 (2.7)	37 (31–38)	2600 (1436–3106)	-	-
C	220 (85.3)	38 (28–42)	2780 (765–4450)	210 (95.5%)	57/220 (25.9%)
D	6 (2.3)	38 (37–40)	3240 (2135–3605)	6 (100%)	-
E	11 (4.3)	37 (34–40)	2990 (2684–4512)	11 (100%)	2/11 (18.2%)
Total	258	38 (28–42)	2697 (765–4512)	227/258 (88%)	60/258 (23.3%)

EA = esophageal atresia.

VACTERL = vertebral defects, anal atresia, cardiac defects, tracheoesophageal fistula, renal anomalies, and limb abnormalities.

anastomosis, and 7/247 (2.8%) patients were treated with an esophageal replacement (all with a gastric tube).

2.3. Anastomotic tension and leak

Anastomotic tension was explicitly documented in 41/247 (16.6%) patients – moderate or high as described in the operative report (Table 2). In those patients, both type A and type B EA had higher rates of tension (4/14, 28.6% and 3/7, 42.9%), when compared with type C (34/220, 15.5%; $p = 0.17$ and $p = 0.03$ respectively). An anastomotic leak occurred in 48/247 (19.4%) patients. Both type A and type B had higher rates of anastomotic leak (7/14, 50% and 4/7, 57.1%), when compared with type C (35/220, 15.9%; $p < 0.01$ and $p < 0.01$ respectively).

2.4. Fundoplication

Anti-reflux surgery (Nissen fundoplication) occurred in 37/247 (15.0%) patients. Both type A and type B had higher rates of Nissen fundoplication (5/14, 35.7% and 2/7, 28.6%) when compared with type C (29/220, 13.2%; $p < 0.05$ and $p = 0.25$ respectively).

2.5. Reoperation

Thirteen of 247 (5.3%) patients underwent reoperations, for a total of 16 reoperations combined. Indications included division of a missed proximal fistula ($n = 5$), anastomotic revision in the setting of an anastomotic leak ($n = 3$), proximal esophagus revision in the setting of an anastomotic leak ($n = 2$), recurrent tracheoesophageal fistula ($n = 2$), excision of tracheal diverticulum ($n = 1$), esophageal perforation during dilatation ($n = 1$), gastric perforation during gap assessment ($n = 1$), and thoracic duct ligation for post-operative chylothorax ($n = 1$).

2.6. Dilatation

The majority (149/247, 60.3%) of patients underwent at least one esophageal dilatation. Of these, 121 (49.0%) underwent two or more dilatations. The median number of dilatations was 4 (interquartile range [IQR] 2–8; range 1–97). In patients with two or more dilatations, the median number of dilatations was 5 (IQR 3–10; range 2–97). The

most common dilatation technique used was bougie alone (92/149, 61.7%), followed by combinations of bougie and balloon dilatation (51/149, 34.2%) (on different days), and balloon alone (6/149, 4.0%). Both type A and B EA patients were more likely to have dilatations (A: 12/14, 85.7%; B: 6/7, 85.7%) when compared with type C (126/220, 57.3%; $p < 0.01$ and $p = 0.01$ respectively). Those patients undergoing balloon dilatation (median 11 months, range 28 days – 8 years 7 months) were younger than those undergoing bougie dilatation (median 2 years, range 24 days – 12 years 11 months; $p < 0.01$).

The dilatation burden, according to age at time of first dilatation, is outlined in Table 3. The eldest patient, based on age at first dilatation, was 9.6 years. Patients who had their first dilatation between 3 and 6 months of age had the highest requirement of dilatation, compared with all other age groups at time of first dilatation.

The three factors found to be significantly associated with an increased dilatation burden were Gross type A and B, anastomosis under tension, and delayed repair:

- (1) Gross type A and B (relative risk [RR] 1.48, 95% CI 1.20–1.82, $p < 0.01$)
- (2) Anastomosis under tension (RR 1.27, 95% CI 1.02–1.58, $p = 0.03$)
- (3) Delayed repair (RR 1.61, 95% CI 1.36–1.90, $p < 0.01$)

Patients who had a primary repair had a lower rate of dilatation (RR 0.65, 95% CI 0.54–0.78 $p < 0.01$) compared with patients with delayed repair or esophageal replacement. However, on logistic regression analysis, only EA type and repair type was found to be statistically significant ($p = 0.01$ and $p = 0.03$).

We found no significantly increased burden of dilatation for the following risk factors:

- (1) VACTERL association
 - 38/58 (65.5%) of patients with VACTERL association required dilatation ($p = 0.09$)
- (2) Cardiac anomalies
 - 47/85 (55.3%) of patients with cardiac anomalies required dilatation ($p = 0.21$)
- (3) Anastomotic leak
 - 30/48 (62.5%) of patients with anastomotic leak required dilatation ($p = 0.43$)

Table 2
Results: key endpoints.

EA type	N (%)	Anastomotic tension (%)	Anastomotic leak (%)	Fundoplication (%)	Dilatation frequency (%)	Median (IQR)
A	14 (5.4)	4/14 (28.6%)	7/14 (50%) †	5/14 (35.7%) *	12/14 (85.7) †	16.5 (7.5–43)
B	7 (2.7)	3/7 (42.9%) *	4/7 (57.1%) **	2/7 (28.6%)	6/7 (85.7) **	12 (5–19)
C	220 (85.3)	34/220 (15.5%) *	35/220 (15.9%) † **	29/220 (13.2%) *	126/220 (57.3) † **	4 (2–6.5)
D	6 (2.3)	-	2/6 (33.3%)	1/6 (16.7%)	4/6 (66.7)	2 (1.2–2.5)
E	11 (4.3)	-	-	-	-	-
Total	258	41/258 (15.9%)	48/258 (18.6%)	37/258 (14.3%)	148/258 (57.4)	4 (2–8)

EA, esophageal atresia.

IQR, interquartile range.

* $p < 0.05$.

† $p < 0.01$.

** $p < 0.01$.

Table 3
Dilatation burden according to age at first dilatation.

Age at first dilatation	N	Median number of dilatations (range)	p-Value ***
<1 month old	2	5.5 (4–7)	0.006
1 month to 3 months old	53	4 (1–16)	0.003
3 months to 6 months	26	11 (1–97)	***
6 months to 1 year old	27	5 (1–40)	0.015
1 year to 2 years old	26	2 (1–18)	0.002
2 years to 5 years old	9	2 (1–4)	0.0005
5 years to 10 years old	6	1.5 (1–2)	0.0004

*** p-Value compared against 3–6 months group, which had the highest requirement for dilatation.

(4) Proton pump inhibitor use

- 74/125 (59.2%) of patients with PPI use required dilatation (p = 0.31)

(5) Esophageal replacement

- 5/7 (71.4%) of patients with esophageal replacement required dilatation (p = 0.71)

2.7. Dilatation complications

There were 13 complications following dilatations, in 1128 dilatation episodes (1.2%); 11 esophageal perforations and two clinically-significant aspiration episodes. All complications were managed conservatively. Perforations were rare for both balloon (6/287, 2.1%) and bougie dilatations (4/841, 0.5%), while one patient had a perforation at the time of guidewire insertion without proceeding to dilatation.

Perforations in both groups were not significantly associated with sex of patient (4/6, 66.7% male in balloon group vs 2/4, 50% male in bougie group, p = 1) nor number of dilatations (median 36.5, IQR 6.5–56.5 versus median 6, IQR 4.5–15, p = 0.13). Age at complication was available for 12/13 episodes (92.3%). Age at balloon dilatation perforation was not significantly different to that of bougie dilatation perforation (median 6.7 months, IQR 2.0–14.9 months vs 12.5 months, range 11.8–18.8 months, p = 0.43). In the balloon perforation group, the patients collectively went on to have 92 further balloon dilatations, and 96 further bougie dilatations; in the bougie perforation group, they went on to have no further balloon dilatations and four further bougie dilatations. Details of dilatation perforations are described in Table 4. Admission details following complications were available for 12/13 episodes (92.3%). In two patients, complication occurred during a prolonged admission for multiple other issues, and the lengths of stay post complication were 13 days and 103 days. The remaining patients who were admitted for management of dilatation complication had a median length of stay of 4 (range 2–16) days. Three patients required stent insertion for treatment of refractory strictures.

3. Discussion

Despite advances in pre- and post-operative care of neonates born with EA, and refinements in operative technique, long-term morbidity is still an issue. Anastomotic stricture and esophageal dysmotility are common complications following EA repair [1, 3, 7, 20–28]. Stricture and dysmotility are important contributors to morbidity, often requiring repeat interventions, as multiple dilatations may be required before a satisfactory result is seen [2, 10]. Approximately 50% of EA patients will require esophageal dilatation(s) [29]. However, it remains possible for surgeons to adopt a ‘watch and wait’ approach. Koivusalo et al. [30] demonstrated that elective dilatation, as a prophylactic measure, does not prevent stricture formation.

In our cohort, patients who presented with suggestive symptoms underwent rigid esophagoscopy to assess for stricture formation, and contrast studies were done at the discretion of the consultant surgeon. This is due to the fact that EA patients face several radiology procedures

Table 4
Details of dilatation perforations.

Year of perforation	Age at perforation (months)	Number of previous dilatations	Number of subsequent dilatations	Type of EA
<i>(a) Balloon dilatation perforations</i>				
2001	2.3	2	4	C
2006	5.6	1	95	A
2007	10.5	11	31	C
2009	7.8	4	35	A
2009	28.3	13	19	A
2015	1.1	0	4	C
<i>(b) Bougie dilatation perforations</i>				
2007	12.5	2	3	C
2014	18.8	17	0	A
2015	Not available	3	0	C
2015	11.8	4	1	B
<i>(c) Guidewire perforation</i>				
2015	5.6	0	13	A

and diagnostic studies, being exposed to an average of 17.4mSV of ionizing radiation in the first 3 years of life (equivalent to 7 years of background radiation) [31]. Therefore, rationalization of diagnostic imaging is important.

Our study identified that more than half of the patients had symptoms of anastomotic strictures or dysmotility preceding dilatation. It is well-established that risk factors for dilatation requirement are multifactorial. Previous studies identified anastomotic tension as a key factor [3]. Serhal et al. [5] identified a stricture rate of 37%, and found that anastomotic tension increased the risk of stricture by a factor of 9. Our study identified anastomotic tension in 16.6% and anastomotic leak in 19.4% of patients. However, stratifying by EA types, we identified that Gross types A and B (i.e. long-gap EA) had higher rates of anastomotic tension and leak when compared with type C.

A study by Salö et al. demonstrated that, in a cohort of 49 EA patients, 39% required at least one esophageal dilatation, and all of these children had their initial dilatation within the first year of life [32]. The authors reported that timing of initial dilatation (< 6 months old versus 6–12 months) was predictive of the need for dilatation beyond the first year (p = 0.03). In our cohort, patients aged 3–6 months at the time of first dilatation had the highest frequency of subsequent dilatations, compared with other age groups.

The presence of GERD has been reported as a key causal factor in the development of anastomotic strictures [5, 12, 33, 34]. Anti-reflux surgery occurs in up to 20% of patients with EA [5, 34, 35]. In our study, types A and B had higher rates of Nissen fundoplication. Overall, anti-reflux surgery occurred in 14.3% of patients, comparable with the literature [5, 34, 36].

Symptoms of GERD may persist, despite anti-reflux therapies, in which case eosinophilic esophagitis needs to be considered. Eosinophilic esophagitis, a chronic, immune-mediated disease, may be associated with strictures following EA repair [8, 9, 37, 38]. Patients with a history of atopy and presence of peripheral eosinophilia are at an increased risk of developing eosinophilic esophagitis [39]. The presence of mucosal eosinophilia is predictive of stricture formation [39]. These strictures tend not to respond well to dilatation, unless the underlying immune mechanism is treated [9]. In our institution, we have not routinely investigated for eosinophilic esophagitis in our EA patients. In light of recent evidence, we will be increasingly focusing upon this.

Balloon and/or bougie dilatations are employed to treat symptomatic post-operative strictures [5]. Lang et al. [40] suggested that balloon dilatation for strictures post-EA repair is more effective and less traumatic when compared with bougie dilatations. Serhal et al. [5] proposed that balloon dilatation is more efficient, as the expansive force is applied uniformly and radially at the stricture site, whereas a bougie exerts a shearing axial force that results in a greater degree of trauma, increasing the risk of perforation.

The reported rate of perforation for balloon dilatations is 0–2.8%, whilst that of bougie dilatation is 8–9% [5, 41]. At our institution, balloon dilatation was preferred for younger patients; however, this did not contribute to the perforation rate. In addition, we determined that perforations from balloon or bougie dilatation did not alter subsequent preferences in dilatation choice for these patients. Younger patients may have more fragile tissues and tighter strictures (smaller caliber of the balloon). However, the use of balloon dilatation may be more effective for patients with an isolated stricture, whilst the use of bougie dilatation may be more effective for patients with dysmotility, due to the mechanics of each technique. Despite the differences, we did not see an age difference in the perforation groups.

The success rate of balloon dilatations is reported to be 63–100% [11, 24, 42–44]. Children with lower grades of dysphagia typically require fewer dilatations, as do younger children [11, 44]. Older children are thought to have denser scarring and potentially longer durations of symptoms, leading to poorer success rates [11]. In addition, the presence of GERD is associated with undergoing more than two dilatations [24].

Bougie dilatation has a variable success rate, ranging from 58 to 96%, depending upon the etiology of the stricture [5, 45]. The majority of our patients underwent at least one esophageal dilatation, with a median number of four. The most common technique at our institution was bougie alone. Proponents of the bougie technique suggest that the operator has greater tactile sensation [46]. Bougies may be used multiple times, so are cost effective, whereas balloon catheters are single use. A rigid bougie may stretch the gastro-esophageal junction, potentially helping with an underlying foregut dysmotility.

In our study, there were 13 complications following dilatations, experienced by 12 patients. This complication rate is similar to the literature, as was the conservative approach to management of these complications [5, 7, 40, 41, 44, 47, 48]. These studies showed a difference in complication rates for balloon and bougie dilatations. In our experience, complications were rare, and there was no difference between techniques. Therefore, the choice of dilatation technique should factor in the above advantages and disadvantages discussed, rather than the risk of complication.

Long-gap EA patients typically undergo dilatations most frequently. In our study, 85.7% of types A and B had requirements for dilatation, and a higher median number of dilatations. Types A and B have increased anastomotic tension, anastomotic leak, fundoplication rates, dilatation requirements and recurrent anastomotic strictures, when compared with type C [7, 33, 49–52]. However, we did not demonstrate that anastomotic leak was a contributing factor in anastomotic stricture development, contrary to other studies [5, 53]. In fact, when logistic regression analysis was performed, the presence of anastomotic tension was not an independent risk factor for dilatation requirement. Rather, it is a factor present in long-gap EA patients, who are the group at risk for dilatation requirement.

Our study is limited by its retrospective nature, albeit utilizing data from a large prospectively maintained database. In turn, our data were reliant upon documentation in the medical records. This is evident in the data collected for our long-gap EA patients, of which seven were documented to have anastomotic tension, as determined by the operating surgeon and documented in the operation note. By definition, it would be expected that these long-gap patients would have a degree of anastomotic tension. However, our findings have showed both long-gap EA and increased anastomotic tension to be risk factors for dilatation burden.

Additionally, it is difficult to objectively define a stricture, and our chosen definition may possibly inflate the stricture rate identified. A child with mild narrowing and mild symptoms may be deemed to have a stricture when, in reality, their symptoms may be secondary to GERD. In light of this, future management of anastomotic stricture may include the Esophageal Anastomotic Stricture Index to provide a more objective means of assessing stricture risk and, therefore, possible dilatation burden [6, 54]. Similarly, anastomotic tension is difficult to

quantify. In the period following the study, intra-operative assessment of tension has been more accurately documented. Assessment of tension is difficult to objectively quantify, based upon surgeon experience, and subjective evaluation and gap length.

Our study has the advantage of data available from one of the world's largest prospective EA databases, the Nate Myers Oesophageal Atresia database, and contributes to our current understanding of dilatation for EA patients in the literature. While anastomotic stricture is reported to range from 9% to 80%, the rate of dilatation in the literature ranges from 39% to 62% [6, 32, 54–59]. Our single-center study, of a large cohort of 258 EA patients, reports an esophageal dilatation rate similar to that in the literature, albeit close to the upper limit. This is likely related to the fact that in our cohort, the decision for dilatation is surgeon-led, rather than imaging-led, which might reduce the threshold for intervention. Our study has also demonstrated that long-gap esophageal atresia and anastomotic tension are significant contributing factors in the development of symptoms of anastomotic strictures and/or dysmotility. Of note, rigid bouginage and balloon dilatation were both safe methods for dilating esophageal strictures, with low incidence of complications.

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Conflicts of interests

None.

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