

Introduction

Pediatric esophageal strictures are most commonly encountered in children following esophageal atresia (EA) repair, or in those who have sustained caustic ingestion injuries. For such children, they are often a long-term source of morbidity, and frequently require multiple interventions to improve their symptoms.

Even in expert hands, strictures at the site of esophageal anastomosis following surgery for EA are not uncommon, cited as symptomatic in more than one third of patients presenting with symptoms to clinic¹ but seen in over 60% of patients on barium swallow assessment². There are a number of factors known to influence the risk of stricture formation, and careful attention to anastomotic integrity, as well as aggressive management of gastroesophageal reflux disease (GERD) have been thought to reduce the risk³. Caustic ingestion is another common indication for stricture dilatation, and a recent study has indicated that it represents an underappreciated global health burden⁴.

Patients with esophageal stricture may require extensive treatment over many years to manage their symptoms, and the majority of patients with stricture will undergo dilatation which has been shown to manage symptoms effectively⁵. Previously, strictures were treated with dilatation with rigid bougie, however management has generally moved forward to balloon dilatation, with the theory that the radial force applied may reduce the risk of esophageal damage and stricture recurrence⁶.

Historically, management of post-operative strictures following EA repair was performed by the operating surgeon. More recently, patients with other disease aetiology such as inflammatory or reflux related strictures have been referred to gastroenterology services and interventional radiology (IR) services have expanded in pediatric practice. These changes have led to a cross-speciality management of esophageal stricture emerging in our centre, as with many others.

Previous studies have analysed single centre outcomes for dilatations in EA^{5,7-10}. However, to our knowledge, no previous paper has studied if there is a difference in practice or outcomes between IR, surgery and gastroenterology.

Methods

Pediatric patients undergoing esophageal dilatation at our centre between April 2014 and December 2018 were identified via retrospective analysis of prospectively maintained databases and clinical coding records. Graded esophageal dilatations were performed with the use of appropriate balloon catheter sizes for the patient's weight, age and stricture severity. The size of the balloon used was up to the discretion of the attending clinician. The balloon catheter was inserted across the stricture after placement of a guidewire under either fluoroscopic or endoscopic guidance and inflated using a radio-opaque contrast material, and obliteration of the "waist" on fluoroscopy was deemed to indicate successful dilatation. In patients where esophageal injury was suspected (i.e. blood noted on the balloon), a post-dilatation contrast study was performed to assess for any evidence of esophageal perforation.

Data were collected on age at presentation, underlying diagnosis, medical speciality, balloon size, increasing size of balloon, interval to subsequent dilation and complications. Data are displayed as median with accompanying range, or mean with standard deviation and are denoted in the text of the results. Appropriate statistical testing of categorical data, mean comparison and distributions were performed using a combination of GraphPad© QuickCalcs and SPSS v23.0 (IBM™).

Results

Over the 4-year study period, thirty-five patients were identified, and all were included with complete records. Patient diagnoses are shown in Table 1, with approximately two thirds having had previous EA repair. Age at presentation was median 18(1-194) months. Single strictures were found in 85%, in the proximal (31%), mid-(37%) and distal (17%) esophagus, with the remaining 15% of patients presenting with multi-level disease. Of note there were two patients presenting with Type-1 achalasia cardia, one has been referred for peroral endoscopic myotomy (POEM), the other has been managed with dilatations alone and is currently symptom free and under routine follow-up.

A total 226 dilatations were performed. Patients underwent a median 3(1-40) dilatations; 26/35(74%) patients required multiple dilatations, including 17/24(71%) patients with EA. Interventional Radiology were responsible for 133 (59%) of dilatations, whilst surgeons performed 58(26%) and 35 (15%) were performed by gastroenterologists.

Each individual patient's dilatations are demonstrated in Figure 1. As is demonstrated, surgeons were more frequently involved in cases of EA vs. other diagnoses (41% dilatations vs. 13%, $p < 0.0001$), and correspondingly, IR and gastroenterology were more frequently involved in non-EA diagnosis patients (IR 65% vs. 51%, $p < 0.05$; gastroenterology 22% vs. 8%, $p < 0.001$). A surgeon most commonly performed the first dilatation in any patient compared to IR or gastroenterology (51% vs. 37% vs. 11%, $\chi^2 = 8.7$, $p < 0.05$). Surgeons were more likely to plan a longer interval before a subsequent dilatation compared to either IR or gastroenterology, 3.8(7.3) months vs. 1.8(3.95) vs. 1.3(1.14) (ANOVA, $p < 0.05$).

There was no significant difference in the balloon size used between specialties (Table 2), however surgeons were more likely to progressively increase the size of balloon as shown in Table 3 ($\chi^2 = 9.55$, $p < 0.01$). In terms of complications, one patient had a radiological evidence of perforation which was successfully managed conservatively with observation alone, therefore our complication rate was 1/226 (0.4%).

Discussion

We believe this to be the first study to compare the practice of pediatric esophageal dilatation across three specialty groups. We have demonstrated that division of this work within our centre is a safe and feasible practice, with a complication rate that is comparable to the published literature¹¹. We demonstrate a variation in practice regarding intervals between dilatations and serial increase in balloon sizes between the services at our centre.

We have identified that surgeons are more likely to increase the balloon size used. The reason behind this is unclear, but capability to manage complications, alongside a more historical acquaintance with the patient and their condition may explain a more 'aggressive' approach in this regard. Comparatively, interventional radiologists may be more hesitant to rapidly upscale the balloon size in surgical patients, especially if they have not previously been involved in their management.

Surgeons were also more likely to leave a longer interval before reattempting dilatation than either of the other two specialist services. We speculate that this may be a selection bias, since patients requiring regular frequent dilatations are commonly referred to the interventional radiology service. Surgeons' use of a slightly smaller balloon size may be explained by the fact that they are commonly the individuals performing the initial dilatations, especially following EA repair (Figure 1), and these patients are then referred to interventional radiology in later life. We would point out that although our case series is smaller than that of other centres reporting from within the United Kingdom^{5,8}, the four year period of data collection denotes a relatively high throughput. We feel that our multi-speciality approach may allow better management of this case volume.

This study bears the limitations of its retrospective nature, and the relatively small size of the cohort may mean that the variability in practice represents variation at an individual level as opposed to between specialities per se. Our single centre data may also not be representative of the wider practice across the specialty; this may be true in fact as the rate of EA patients undergoing multiple dilatations exceeded 70%, compared to approximately half the patient series reported in other centres^{5,7-9}, suggesting a difference in threshold for intervention. The heterogeneous nature of this cohort may also confound our findings, as different management plans in terms of frequency and size of dilatations may be instigated for strictures of different aetiologies.

Esophageal strictures are a source of significant morbidity, and as patients often require repeated procedures, timely management is critical. Our study suggests that such a multi-disciplinary approach can be adopted successfully, with similar practices demonstrated between specialist teams and an

overall complication rate in keeping with the published literature. We hope that this work will expand upon the existing knowledge available, and help other centres to consider a cross-specialty approach in these patients.

Disclosure Statement

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