FLUOROSCOPIC BALLOON DILATATION FOR ANASTOMOTIC STRICTURES IN PATIENTS WITH ESOPHAGEAL ATRESIA

A FIFTEEN YEAR SINGLE CENTRE UK EXPERIENCE

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## Abstract

### Aim of the Study

To assess the safety and effectiveness of fluoroscopic balloon dilatation (FBD) in children with esophageal anastomotic stricture after surgical repair of esophageal atresia.

### Methods

All patients undergoing surgery for esophageal atresia and requiring dilatation(s) during a consecutive 15 year period [ April 2000 - September 2014 ] were analyzed. Dilatations were performed as day case procedures under general anesthesia using a radial force generating balloon device ( Boston Scientific Corporation ) by surgeons. Outcomes assessed included – (1) the number of dilatations / patient, (2) effectiveness and (3) need for surgery and (4) complications.

### Results

One hundred thirty seven patients underwent 625 FBD sessions (median 3 dilations per patient; range 1–24 dilatations). Median age at 1st FBD was 0.74 years (range 0.05–16.1 years). Balloon catheter sizes ranged from 6 mm – 20 mm. FBD yielded excellent results in 99 patients (74 %), while 17 cases (13 %) had mild ongoing dysphagia / dysmotility. Ten patients (7%) required further dilatation(s) to control symptoms. No patient(s) required esophageal stenting. Five cases required G-tube feeds as a result of oral aversion behavior – all of these cases were complex / VACTERL patients. Only 1 minor ‘radiological leak’ occurred after a dilatation session and this did not require surgical intervention. A single patient (‘long gap’ EA-TEF) with severe neurological impairment having multiple dilatations and stricture resection ultimately required esophageal replacement. Anti-reflux surgery was performed in 36 patients (26 %) for medical therapy resistant GER.

### Conclusion

FBD for anastomotic stricture(s) following esophageal atresia repair achieved very good outcomes for the majority of EA-TEF patients. The procedure can be accomplished safely as indicated by the low complication rate herein reported. Although some children may require more than one dilatation session prompt relief of symptoms can be achieved with a vigilant care programme co-ordinated by a multidisciplinary specialist EA TEF team.

## Introduction

Esophageal atresia (EA) occurs in 1 in 2500 births [1]. Survival today is generally excellent though surgery for EA is associated with well recognised morbidity(s) - anastomotic stricture, leak, foregut dysmotility, recurrent fistula, gastroesophageal reflux (GER) disease and airway malacia [1-4]. It is estimated that up to 50 % of all EA patients will acquire swallowing problems linked to stricture with the subsequent risk(s) of dysphagia and aspiration [2,5]. After-care programmes have traditionally advocated esophageal dilatation with rigid or flexible bougies. It is postulated that such procedures may be associated with a higher rate of esophageal injury resulting from the ‘shearing forces‘ created giving further scarring and worsening strictures [6]. Balloon dilatation is considered to be a superior technique as the ‘radial dilating forces ‘ from the device are theoretically less likely to produce esophageal damage [5]. Balloon dilatation can be accomplished under fluoroscopic imaging with patients often managed by interventional radiology services [7-11]. To this end only very few pediatric surgical centers have accumulated ‘surgeon experience’ with the technique [5,12-14]. This paper herein reports a 15 year single center study undertaken by a multidisciplinary surgical service caring for EA TEF patients .

## Methods

All patients undergoing surgery for EA at Alder Hey Children’s Hospital, Liverpool UK during the 15 year period, April 2000 - September 2014 were identified from an EA TEF clinical database. Cases requiring FBD for anastomotic stricture(s) after EA repair were then detailed. Strictures were suspected by ‘symptom reporting’ by patients, parents or ‘care-givers’. Water soluble upper GI contrast studies were deployed occasionally for some patients ( requested by parents and care-givers ) to aid decision-making from clinics and / or outreach ambulatory services. All dilatation sessions were performed by pediatric surgeons. As routine - flexible upper GI endoscopy ( Olympus Corporation , UK ) was undertaken on each patient prior to dilatation to exclude food bolus, foreign body or debris entrapment in the esophagus. Appropriate balloon catheter size(s) ( 6mm – 20 mm ) were then selected by the attending surgeon after endoscopy taking account of the severity of stricture, patient age and body weight. A balloon catheter was inserted into the esophagus over a soft flexible fine guide wire positioned across the length of the stricture. With the “ radial-force “ generating balloon device ( Boston Scientific Corporation ) using fluoroscopy control - stricture(s) were dilated. At each dilatation the balloon was inflated with dilute radio-opaque contrast (Omnipaque, GE Healthcare UK) for periods of up to 60 seconds and then deflated gradually. Disappearance of the “waist” or “hourglass” deformity indicated successful dilatation – Figure 1. This procedure was repeated up to 2 – 3 times per patient at each session. An ‘on table ‘ post dilatation esophagogram study was performed by instillation of contrast through a working channel of the balloon device to exclude ‘leak’. Postoperatively feeding was allowed on the ward immediately after recovery from anaesthesia. Outpatient clinic attendance and medical chart review(s) were analyzed to evaluate outcomes. Data recorded for each patient included - ( 1 ) the number and frequency of dilatations / patient, ( 2 ) clinical effectiveness / ‘ symptom relief ‘ ( 3 ) need for surgery and ( 4 ) complications.

## Results

One hundred thirty seven patients underwent a total of 625 FBD sessions over a 15 year period. The median number of dilatations required was 3 per patient (range 1–24 dilatations) - Figure 2. Median age at 1st FBD was 0.74 years (range 0.05–16.1 years). FBD yielded excellent results with complete resolution of symptoms in 99 patients (74 %), while 17 patients (13 %) had mild ongoing dysphagia / dysmotility disturbance. These children were able to tolerate normal diet and managed to control their symptoms by ensuring to drink lots of water / fluids at meal times. Ten patients (7 %) required further dilatation(s) sessions to control troublesome symptoms. A single FBD session was highly sufficient in 28 patients (21 %) whereas ‘symptom recurrence’ required repeat dilatations in 109 children (79 %). Morbidity in the study was minimal with only 1 single case of oesophageal perforation – a ‘radiological leak’ - (0.7 % per patient; 0.16 % per dilatation session) which was managed conservatively. Post operatively 1 patient experienced a severe desaturation episode in the recovery ward who was later found to have severe tracheomalacia that required aortopexy. Despite effective dilatations, five patients remain mainly tube fed (4 gastrostomies, 1 jejunostomy). All of these children were more complex cases / phenotypes - 1 cerebral palsy and VACTERL complex, 1 ‘developmental delay’, 2 VACTERL syndrome and 1 patient with short bowel syndrome – Figure 3. A single adolescent patient with ‘long gap’ oesophageal atresia and severe neurological impairment / post infective viral brain injury ultimately required oesophageal replacement after multiple dilatations and failed stricture resection. Antireflux surgery was undertaken in 36 patients (26 %) for medical therapy resistant GER. There were three deaths during follow up - All were unrelated to OA and FBD – ( 2 severe co-existent congenital heart disease and 1 CVL line sepsis). The median follow up period in the study was 4.9 years (range 0.14 – 15.5 years). During this 15 year era the pediatric surgical unit performed 181 esophageal atresia repairs – approximately 12 index cases per annum.

## Discussion

Early reports describing balloon dilatation for esophageal strictures in children date back to 1984 [15,16]. Symptom relief after first dilatation is reported to occur in 15% – 59 % cases [5,8,10,17,18] comparable with the good success rates in this present study. The median number of dilatations required per patient ( reportedly 3 sessions ) , gave an overall success rate of 76 % / index case similar to the contemporary literature [5,7-10,17]. The findings from this single center UK study reaffirms that FBD is a very safe and effective technique for management of anastomotic strictures following EA repair. The most serious complication(s) of dilatation procedures is esophageal perforation with the reported risk(s) in the literature quoting 0 %– 10 % incidence [8,11-13]. Herein we reported only a single case - a perforation / ‘radiological leak ‘ after FBD giving a perforation rate of 0.16 % per procedure.

It is estimated that up to 70 % of infants born with EA TEF will have co-existent GER disease considered to be a significant contributing factor in the pathogenesis of postoperative strictures [2,9,19]. It is well known that most strictures can resolve with fewer dilatations necessary after antireflux surgery [2] and also that fewer dilatations are required in patients without GERD [17]. In our series antireflux surgery was performed in only 36 patients (26 %) for medical therapy refractory GER where we reserve a ‘high threshold’ for electing to perform fundoplication in EA patients with well recognised foregut dysmotility. These outcome metrics are very similar with findings as reported by Chittmittrapap [2] (26 %) and Said. [9]

FBD is therefore highly effective although it must be recognized that some patients will require further sessions ( decreasing in overall frequency as adolescence years advance ) , whilst others may remain mainly tube-fed due to the complexity of their associated conditions / aversive oral behaviour. In Liverpool vigilant after-care in a specialist MDT EA monthly clinic has greatly facilitated active surveillance of index patients also considerably reducing ‘out of hours’ emergent hospital admission for emergent dilatation(s). Here we practice a selective policy for dilatation for ‘symptomatic strictures’ as advocated by Koivusalo et al in Finland [20,21]. To add here - ‘symptom threshold’ for booking (re) dilatation session(s) in our practice is swift in a strenuous effort to offset emergency admission from bolus obstruction dysphagia. [2,5,17,22]. To the best of our knowledge this is the largest single center series reporting successful deployment of FBD to treat esophageal anastomotic strictures after OA repair. In closing - the technique is readily learned by pediatric surgeons-in-training and we believe it is a crucial skill-set to acquire for any surgeon(s) wishing to develop a subspeciality interest in the care of babies born with esophageal atresia.

References

[1] Goyal A, Jones MO, Couriel JM, Losty PD. Oesophageal atresia and tracheo-oesosphageal fistula. Arch Dis Child Fetal Neonatal Ed 2006; 91: F381-4

[2] Chittmittrapap S, Spitz L, Kiely EM, et al: Anastomotic stricture following repair of esophageal atresia. J Pediatr Surg 1990;25:508-511

[3] Chittmittrapap S, Spitz L, Kiely EM, et al: Anastomotic leakage following surgery for esophageal atresia. J Pediatr Surg 1992;27:29-32

[4] Spitz L: Oesophageal atresia. Orphanet J Rare Dis 2007;2:24

[5] Lang T, Hummer HP, Behrens R: Balloon dilation is preferable to bougienage in children with esophageal atresia. Endoscopy 2001;33:329-335

[6] Serhal L, Gottrand F, Sfeir R, et al: Anastomotic stricture after surgical repair of esophageal atresia: frequency, risk factors, and efficacy of esophageal bougie dilatations. J Pediatr Surg 2010;45:1459-1462

[7] Johnsen A, Jensen LI, Mauritzen K: Balloon-dilatation of esophageal strictures in children. Pediatr Radiol 1986;16:388-391

[8] Ko HK, Shin JH, Song HY, et al: Balloon dilation of anastomotic strictures secondary to surgical repair of esophageal atresia in a pediatric population: long-term results. J Vasc Interv Radiol 2006;17:1327-1333

[9] Said M, Mekki M, Golli M, et al: Balloon dilatation of anastomotic strictures secondary to surgical repair of oesophageal atresia. Br J Radiol 2003;76:26-31

[10] Thyoka M, Barnacle A, Chippington S, et al: Fluoroscopic balloon dilation of esophageal atresia anastomotic strictures in children and young adults: single-center study of 103 consecutive patients from 1999 to 2011. Radiology 2014;271:596-601

[11] Fasulakis S, Andronikou S: Balloon dilatation in children for oesophageal strictures other than those due to primary repair of oesophageal atresia, interposition or restrictive fundoplication. Pediatr Radiol 2003;33:682-687

[12] Tam PK, Sprigg A, Cudmore RE, et al: Endoscopy-guided balloon dilatation of esophageal strictures and anastomotic strictures after esophageal replacement in children. J Pediatr Surg 1991;26:1101-1103

[13] Sandgren K, Malmfors G: Balloon dilatation of oesophageal strictures in children. Eur J Pediatr Surg 1998;8:9-11

[14] Shah MD, Berman WF: Endoscopic balloon dilation of esophageal strictures in children. Gastrointest Endosc 1993;39:153-156

[15] Goldthorn JF, Ball WS,Jr, Wilkinson LG, et al: Esophageal strictures in children: treatment by serial balloon catheter dilatation. Radiology 1984;153:655-658

[16] Ball WS, Strife JL, Rosenkrantz J, et al: Esophageal strictures in children. Treatment by balloon dilatation. Radiology 1984;150:263-264

[17] Antoniou D, Soutis M, Christopoulos-Geroulanos G: Anastomotic strictures following esophageal atresia repair: a 20-year experience with endoscopic balloon dilatation. J Pediatr Gastroenterol Nutr 2010;51:464-467

[18] Lan LC, Wong KK, Lin SC, et al: Endoscopic balloon dilatation of esophageal strictures in infants and children: 17 years' experience and a literature review. J Pediatr Surg 2003;38:1712-1715

[19] Tovar JA, Diez Pardo JA, Murcia J, et al: Ambulatory 24-hour manometric and pH metric evidence of permanent impairment of clearance capacity in patients with esophageal atresia. J Pediatr Surg 1995;30:1224-1231

[20] Koivusalo A, Turunen P, Rintala RJ, et al: Is routine dilatation after repair of esophageal atresia with distal fistula better than dilatation when symptoms arise? Comparison of results of two European pediatric surgical centers. J Pediatr Surg 2004;39:1643-1647

[21] Koivusalo A, Pakarinen MP, Rintala RJ: Anastomotic dilatation after repair of esophageal atresia with distal fistula. Comparison of results after routine versus selective dilatation. Dis Esophagus 2009;22:190-194

[22] Allmendinger N, Hallisey MJ, Markowitz SK, et al: Balloon dilation of esophageal strictures in children. J Pediatr Surg 1996;31:334-336

Legends to Figure(s)

Figure 1 – Fluoroscopy imaging showing the technique of esophageal balloon dilatation. The ‘ waist ’ deformity defines the area of stricture in the esophagus. Absence of ‘ waisting ’ with balloon inflation indicates successful dilatation.

Figure 2 – Bar chart plot showing number of dilatation sessions required per EA patient.

Figure 3 – Bar chart plot – Defining outcome metrics in EA study cohort.

**Figure 1**







