



Anastomotic stricture after surgical repair of esophageal atresia: frequency, risk factors, and efficacy of esophageal bougie dilatations

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Received 19 August 2009; revised 20 October 2009; accepted 9 November 2009

Key words:

Anastomotic stricture;
Esophageal atresia;
Bougie dilatations;
Endoscopy;
Children

Abstract

Aims: The aim of this study was to evaluate the frequency and risk factors of postoperative anastomotic stricture, and the efficacy and complications of esophageal bougie dilatations for symptomatic anastomotic stricture in a population of children with esophageal atresia.

Patients and Methods: The medical records of 62 children operated on for esophageal atresia type III (Ladd and Gross) over a 5-year period were retrospectively reviewed.

Results: Anastomotic stricture developed in 23 (37%) of patients. Anastomotic tension during primary repair of esophageal atresia was associated with subsequent stricture formation ($P < .05$). Patients required esophageal dilation at a mean age of 149 days (range, 30–600 days). Stricture resolution occurred after a mean of 3.2 dilatations per patient (range, 1–7). Dilation was successful in 87% of patients. Three patients continued to present mild ($n = 1$) to severe ($n = 2$) dysphagia, mainly related to esophageal dysmotility. No complications were observed during or after the dilatation sessions.

Conclusions: Anastomotic stricture, secondary to the surgical treatment of esophageal atresia, remains a frequent complication in patients with esophageal atresia. Esophageal dilation with Savary-Gilliard bougies is a safe and effective procedure in the management of strictures.

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Anastomotic stricture is the main complication after surgical repair of esophageal atresia (EA). It occurs in 18% to 50% of patients with EA that has been surgically repaired [1-3]. Several predisposing factors have been implicated in the pathogenesis of the anastomotic stricture including the nature of the suture materials used to perform the anastomosis, anastomotic tension, anastomotic leakage, fistula, gap length, and gastroesophageal reflux [3-11]. Recent advances in surgery and neonatal care have provided a better prognosis for EA [3,5,7,8]. Indeed, the survival rate of infants born with EA, with or without tracheoesophageal fistula, has dramatically improved over the last decade, increasing from 80% to more than 95% [9]. However, anastomotic stricture remains a frequent complication after the surgical repair of EA. Esophageal balloon or bougie dilatations remain the treatment of choice for symptomatic esophageal strictures.

The purpose of this study was to evaluate the primary surgical repair of Ladd-Gross classification EA types III or V in children at birth. The frequency of and risk factors associated with postoperative anastomotic stricture, and the efficacy and complications of esophageal dilatations for symptomatic anastomotic strictures were analyzed.

1. Patients and methods

1.1. Patients

A retrospective longitudinal study was carried out on children with EA who were operated on between January 2000 and December 2005. This allowed a homogenous population with more than 3 years of follow-up to be studied. The study population included patients undergoing primary repair of EA at birth. The characteristics of these patients were collected from patient charts, operative reports, endoscopic reports, and office notes and entered into a database. Of the 73 patients, 11 were excluded from the analysis: 9 were the Ladd-Gross classification EA type I, and 2 patients suffered early mortality from major cardiac problems or severe prematurity. Of the 62 patients included in the analysis, 61 were Ladd-Gross classification EA type III (ie, EA with distal esophagotracheal fistula) and 1 was Ladd-Gross classification EA type V (ie, EA with proximal and distal esophagotracheal fistulas). The repair of EA was performed within the first day of life in 22 patients, the second day of life in 37 patients, and the third day of life in the remaining 3 patients.

Data regarding sex, birth weight, gestational age, anastomotic tension during primary repair of EA (evaluated by the surgeon), duration of transanastomotic and thoracic tubes, and early postoperative complications, especially anastomotic leakage and recurrent fistula, were noted. The presence of symptoms suggesting gastroesophageal reflux (GER), positive 24-hour pH monitoring, and/or endoscopic signs suggestive of GER were noted. In this study, the

symptoms that revealed anastomotic stenosis included swallowing difficulties (dysphagia, vomiting, cough, poor or slow feeding), foreign body obstruction (including food bolus), recurrent respiratory tract infections, and/or poor weight gain. All patients presenting with at least one symptom suggestive of anastomotic stenosis underwent contrast x-ray to confirm the stricture and identify its location and severity. When stenosis was diagnosed, esophagoscopy and anastomotic dilatation were carried out. No routine prophylactic dilatation was conducted. The characteristics of patients presenting with anastomotic stenosis were compared with those of patients without anastomotic stenosis. Particular attention was given to the efficacy and number of dilatations per patient, the interval between surgical repair of EA and the first esophageal dilatation for symptomatic stenosis, as well as dilatation-related complications.

1.2. Esophageal dilatation

All dilatations were performed with Savary-Gilliard polyvinyl bougies (5, 7, 9, 11, 12.8, and 15 mm) in an operating room on patients under general anesthesia with endotracheal intubation. The choice of endoscope was dependent upon the weight of the child and the esophageal lumen at the level of the stricture. Four types of endoscope were used (Olympus NP30, XP20, and P140 Olympus, Rungis, France and Pentax EG1870K Pentax, Argenteuil, France). Once the stenosis was visualized endoscopically, a flexible tipped guide wire was passed via the side channel of the endoscope into the esophageal lumen; afterward, the endoscope was withdrawn, leaving the guide wire in place. The position of the guide wire was then controlled by fluoroscopy. The dilator was passed over the guide wire and placed across the stricture, and the probe was left in place for at least 1 minute. During each dilatation session, serial increases in diameter were performed that varied between 5 and 12.5 mm. After dilator retraction, an endoscopic control was systematically conducted to evaluate the results of dilatations and the status of the esophageal mucosa. Fluoroscopy was used to monitor the dilator position. The presence of esophagitis led to optimization of antireflux treatment (initiation of or increasing treatment with proton pump inhibitors). The objective of each dilatation session was to break the stricture or to increase the esophageal diameter, thereby allowing the child to swallow without dysphagia. This procedure was followed by a 24-hour stay in hospital for clinical observation. A chest x-ray was systematically carried out 6 hours after the procedure to look for complications, especially esophageal perforation.

Dilatations were considered successful when adequate lumen was achieved with complete relief of dysphagia and weight gain or growth. The frequency and interval between dilatation sessions depended upon the evolution of symptoms suggestive of esophageal stricture and endoscopy data concerning esophageal stricture.

1.3. Statistical analysis

Descriptive analysis was performed to check and summarize the data. Bivariate analysis was performed to determine the predictive factors of stricture formation. Comparisons of frequencies were examined using the χ^2 test or the Fisher's Exact test when the χ^2 test was not appropriate, and comparisons of means were performed using the nonparametric Wilcoxon's test. Parameters that were significant ($P < .05$) after bivariate analysis were introduced in a stepwise logistic regression using SPSS (Chicago, IL). Adjusted odds ratio and 95% confidence intervals were then calculated for the parameters significantly associated with the formation of esophageal stricture.

2. Results

The anastomotic stricture rate after surgical repair of EA in our series of patients was 37% (23 of 62 patients). The analysis of risk factors for stricture formation showed a significant relationship between stricture formation and prematurity (born before 37 weeks of gestation) ($\chi^2 = 4.45$, $P = .035$), VACTERL syndrome (Fisher's Exact test, $P = .013$), GER ($\chi^2 = 4.79$, $P = .029$), anastomotic tension (Fisher's Exact test, $P = .024$), and anastomotic leakage (Fisher's Exact test, $P = .043$). In contrast, the stricture rate was unaffected by sex, intrauterine growth retardation, tracheomalacia, duration of intubations and use of curare per operation, and duration of transanastomotic and transthoracic drains. Multivariate analysis revealed that only anastomotic tension was significant (odds ratio: 9.23 [2.19-37.87], $P < .0024$).

Prokinetic and/or proton pump inhibitor therapy was provided to 77% of patients (48 patients) who presented with clinical symptoms of GER confirmed by 24-hour esophageal pH monitoring and/or endoscopy. Twenty-six patients (42%) presented with peptic esophagitis. Esophagitis was noted in 61% of patients with stricture compared with 31% of patients without stricture ($P < .05$). In the 13 patients for whom treatment failed, GER was further treated by surgical procedures (Nissen fundoplication in 11 patients and total esophagogastric dissociation, namely, the Bianchi procedure, in 3 patients). Surgical management of GER was performed in 34% of patients with anastomotic stenosis compared with 13% of patients without anastomotic stenosis ($P < .05$). In 2 patients, EA was associated with congenital esophageal stricture that required surgical management in 1 patient and 3 sessions of esophageal dilatations in the other patient.

The delay between surgical treatment of patients with EA and the first esophageal dilatation varied from 30 to 600 days (mean, 149 days). Stricture resolution occurred after a mean of 3.2 dilatations per patient (range, 1-7) over an average period of 7 months (range, 0-55 months). Four patients (17%) required a single dilatation; 5 (21%) needed 4 or more

dilatations. Local application of mitomycin C was performed in 1 patient who required 7 dilatation sessions. The mean period of follow-up from the time of the last bougie dilatation was 47 months (varying from 8 to 86 months). Dilatation was successful in 87% of patients (20/23 patients). Esophageal dilatation was unsuccessful in 3 patients, who continued to present with mild ($n = 1$) to severe ($n = 2$) dysphagia mainly related to esophageal dysmotility. These patients underwent total esophagogastric dissociation (Bianchi procedure) because of the presence of gastric pull-up in 2 patients and small stomach in the third patient. Among these patients, 2 received enteral nutrition via percutaneous gastrostomy; and the other patient received oral nutrition. No complications related to esophageal dilatation such as infections, mediastinitis, or perforations were observed during or after the dilatation sessions.

3. Discussion

Despite the recent refinements of operative techniques and improvements in preoperative and postoperative management of the newborn infant, anastomotic stricture after repair of EA remains frequent and is in the same range as that observed in the early 1980s [3]. Indeed, in the present study, the anastomotic stricture rate was 37%, close to the 40% stricture rate that we reported in our center between 1990 and 1995 [6].

As noted in previous studies, our study confirms that anastomotic tension, which is highly correlated with gap length, plays an important role in the development of subsequent stricture [6,11-13]. Brown and Tam [13] showed a significant relationship between gap length and stricture formation (44% stricture formation in a group of infants with long gaps [>3 cm] vs 17% in the other group [gap length <1 cm]). In our multivariate analysis, the only independent factor was the anastomotic tension that increased the risk of esophageal stenosis by a factor of 9. Although its definition is subjective, in our hands, anastomotic tension seems to be a better predictor of stricture formation than length of gap per se in patients with EA types III and V.

Gastroesophageal reflux has been demonstrated to be a significant factor in the formation of postoperative stricture and its recurrence [3,14,15]. However, in our study, multivariate analysis showed the absence of an association between GER and subsequent stricture formation. This discrepancy may be explained by the recent prescription of a systematic proton pump inhibitor treatment in our population.

In our series, esophageal dilatation was used only in symptomatic patients and was successful in 87% of patients. This finding is compatible with that reported in the literature in which the success rate of bougie dilatation varies from 58% to 96% depending upon stricture etiology [6,16]. Esophageal strictures can be dilated effectively with a bougie or balloon catheter [6]. Dilatation with a balloon is

theoretically more efficient because the expansive force is applied uniformly and radially at the site of the stricture, whereas a bougie exerts a shearing axial force that results in a greater degree of trauma and thereby increases the risk of perforation [17].

No prospective study on the efficacy and safety of Savary-Gilliard bougie dilatation compared with balloon dilatation in homogenous populations has been reported. However, in a retrospective study, Lang et al [18] showed that balloon esophageal dilatation is superior to bougie dilatation in safety and efficacy. For uncomplicated esophageal strictures, the perforation rates for balloon dilatation and bougienage are 0% to 2.8% and 8% to 9%, respectively [17-19]. In the present study, the complication rate for bougienage dilation was 0%.

These data suggest that, whatever the technique is (balloon or Savary-Gilliard bougie), esophageal dilatation procedures require a trained operator to reduce the risk factors and complications after esophageal dilatations [6].

References

- [1] Chetcuti P, Phelan PD. Gastrointestinal morbidity and growth after repair of oesophageal atresia and tracheo-oesophageal fistula. *Arch Dis Child* 1993;68:163-6.
- [2] Nambirajan L, Rintala RJ, Losty PD, et al. The value of early postoperative oesophagography following repair of oesophageal atresia. *Pediatr Surg Int* 1998;13:76-8.
- [3] Chittmitrapap S, Spitz L, Kiely EM, et al. Anastomotic stricture following repair of esophageal atresia. *J Pediatr Surg* 1990;25:508-11.
- [4] Louhimo I, Lindahl H. Esophageal atresia: primary results of 500 consecutively treated patients. *J Pediatr Surg* 1983;18:217-29.
- [5] Tsai JY, Berkery L, Wesson DE, et al. Esophageal atresia and tracheoesophageal fistula: surgical experience over two decades. *Ann Thorac Surg* 1997;64:778-83.
- [6] Michaud L, Guimber D, Sfeir R, et al. Anastomotic stenosis after surgical treatment of esophageal atresia: frequency, risk factors and effectiveness of esophageal dilatations [in French]. *Arch Pediatr* 2001; 8:268-74.
- [7] Touloukian RJ, Seashore JH. Thirty-five-year institutional experience with end-to-side repair for esophageal atresia. *Arch Surg* 2004;139: 371-4.
- [8] Spitz L. Esophageal atresia: past, present, and future. *J Pediatr Surg* 1996;31:19-25.
- [9] Konkin DE, O'Hali WA, Webber EM, et al. Outcomes in esophageal atresia and tracheoesophageal fistula. *J Pediatr Surg* 2003; 38:1726-9.
- [10] Spitz L, Kiely E, Brereton RJ. Esophageal atresia: five year experience with 148 cases. *J Pediatr Surg* 1987;22:103-8.
- [11] Jolley SG, Johnson DG, Roberts CC, et al. Patterns of gastroesophageal reflux in children following repair of esophageal atresia and distal tracheoesophageal fistula. *J Pediatr Surg* 1980;15: 857-62.
- [12] Boyle Jr EM, Irwin DE, Foker EJ. Primary repair of ultra-long-gap esophageal atresia: results without a lengthening procedure. *Ann Thorac Surg* 1994;57:576-9.
- [13] Brown AK, Tam PKH. Measurement of gap length in esophageal atresia: a simple predication of outcome. *J Am Coll Surg* 1996;182: 41-5.
- [14] Bergmeijer JH, Hazebroek FWJ. Prospective medical and surgical treatment of gastroesophageal reflux in esophageal atresia. *J Am Coll Surg* 1998;187:153-7.
- [15] Pieretti R, Shandling B, Stephens CA. Resistant esophageal stenosis associated with reflux after repair of esophageal atresia: a therapeutic approach. *J Pediatr Surg* 1974;9:355-7.
- [16] Weintraub JL, Eubig J. Balloon catheter dilatation of benign esophageal strictures in children. *J Vasc Interv Radiol* 2006;17: 831-5.
- [17] Kim IO, Yeon KM, Kim WS, et al. Perforation complicating balloon dilation of esophageal strictures in infants and children. *Radiology* 1993;189:741-4.
- [18] Lang T, Hümmel HP, Behrens R. Balloon dilation is preferable to bougienage in children with esophageal atresia. *Endoscopy* 2001;33: 329-35.
- [19] LaBerge JM, Kerlan Jr RK, Pogany AC, et al. Esophageal rupture: complication of balloon dilatation. *Radiology* 1985;157:56-60.