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Endoscopic biliary stent placement for anastomotic stricture following esophageal atresia repair in infant

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ABSTRACT

Esophageal atresia is the most common congenital anomaly of the esophagus. Treatment optimally includes primary tension-free anastomosis of the upper and lower aspects of the esophagus. Distance between the upper and lower esophagus predisposes the anastomosis to tension, increasing the risk of anastomotic stricture. Esophageal stricture has historically been managed with repeated endoscopic balloon dilations. Multiple failed attempts at endoscopic balloon dilation requires surgical revision of the anastomosis. Stenting of esophageal stricture has been used in the adult population but has not been described for children. This case report demonstrates the placement of biliary stents in a 2-month-old infant with esophageal stricture following repair of a long gap Type C tracheoesophageal fistula. Successful stenting procedure averted the need for surgical revision.

1. Introduction

Anastomotic stricture after repair of esophageal atresia is a common complication. Endoscopic balloon dilation is first-line treatment for esophageal strictures, with most patients requiring 3 dilations [1]. A systematic review that analyzed outcomes for balloon dilation in children with esophageal atresia showed reported success rates from 70-100%, with approximately 3 dilations per patient [2–7]. Strictures unrelieved by endoscopic balloon dilation require surgical revision of the anastomosis. Endoscopic stenting has potential to provide an alternative to surgical revision for patients who have failed multiple balloon dilations to decrease the need for surgical revision.

Endoscopic biliary devices can be useful for dilation and stenting otherwise nontraversable strictures [8]. Esophageal stenting has been used in the adult population for strictures associated with esophageal disease. Use of biliary stents for esophageal stricture has only been described once in an older pediatric population [9]. Given the recidivism seen with balloon dilations of anastomotic strictures, endoscopic stenting could prove to be an additional and useful step in management of complex anastomotic strictures prior to surgical revision.

2. Case report

A 38-week gestation 2595-g newborn was found to have a Type C tracheoesophageal fistula with a 2.5-cm long gap atresia. The patient underwent operative repair on day of life 2 with primary end-to-end anastomosis. The repair was difficult and required revision in the operating room; the anastomosis was under some tension at the conclusion of the procedure due to the long gap atresia. On post-operative day 7, the patient underwent esophogram, which resulted in iatrogenic perforation through the anastomosis. The esophagram was performed in radiology without surgery present. While manipulating the nasogastric tube, that had been left in place at the initial operation, to obtain imaging the tube perforated the right side of the anastomosis. This was confirmed at the time on imaging with contrast extravasation seen into the right chest and later in the chest tube. Repeat imaging one week later was negative for anastomotic leak, but revealed mid-esophageal stricture that persisted on imaging approximately 1 month post op (Fig. 1). The patient demonstrated feeding difficulty; therefore, he underwent serial balloon dilations (up to 8 mm) of the stricture, which failed to improve symptoms.

At 2 months of age, the patient continued to have feeding difficulties without relief despite serial endoscopic balloon dilations. An 8-mm by 4-cm Viabil biliary stent (Gore Creative Technologies; Flagstaff, AZ) was placed across the stricture, approximately 2 mm above the level of the narrowed segment (Figs. 2 and 3). The stent was dilated with a 6-mm balloon and appropriate position was confirmed with fluoroscopy. Following stent placement, the patient was able to tolerate oral feeding. Since initial placement at 2 months of age, the stent has been

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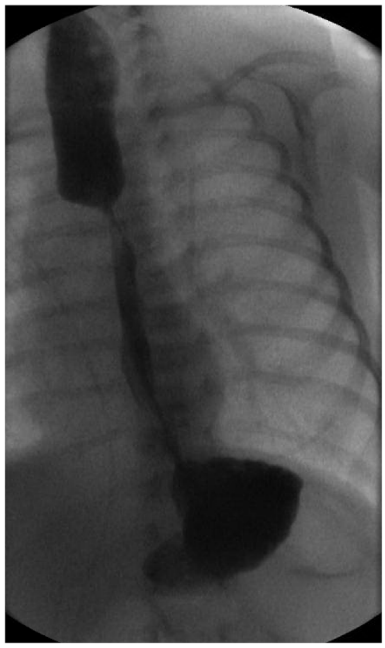

Fig. 1. Anastomotic stricture following tracheoesophageal fistula repair, Postop Day 29.

exchanged 5 times, with stent in place ranging from 25-59 days (average 50 days). The stents were left in for an average of 6–8 weeks due to the theoretical scar remodeling that happens during this time, with the goal of minimizing residual scar tissue at removal. The Viabil stent is capable of being permanent but not needed for our purposes. The stents are retrieved endoscopically with graspers and at the time of removal the stricture was evaluated for replacement of the stent or observation based on the results. At 7 months old, the stent was removed and left out for a period of 5 months. At 12 months of age, the patient had some recurrent issues with oral tolerance and repeat imaging showed continued stricture, so a new 10-mm by 4-cm Viabil biliary stent was placed. Thereafter, this stent has been replaced one time. At 16 months of age, repeat evaluation of the anastomotic stricture showed that it had resolved. The patient continues to do well, but now is having issues from a stricture at the GE junction related to his reflux disease. At this time, he has not required surgical revision of his anastomotic stricture. On most recent imaging, the esophageal stricture has completely resolved.

3. Discussion

Esophageal atresia is the most common congenital anomaly of the esophagus. This anomaly is associated with tracheoesophageal fistula with an incidence of 1 in 3500 live births [10]. Tracheoesophageal fistula is managed by surgical ligation of the fistula and optimally primary anastomosis of the esophageal segments. Anastomotic stricture is a common complication following surgical repair [11]. Factors associated with stricture formation include poor surgical technique, significant tension at the anastomosis, long gap atresia, gastroesophageal reflux disease, and anastomotic leak. In a study of 50 patients with tracheoesophageal fistula with esophageal atresia, the length of the esophageal atresia was correlated with the incidence of postoperative stricture – 100% patient with atresia length greater than 3.5 cm experienced postoperative stricture, and 75% of patients with atresia length between 2 cm and 3.5 cm experienced postoperative stricture [12]. The patient had a 2.5-cm atresia at the time of initial operation, putting the anastomosis at high risk of stricture. Although many could argue that atresia of such length should not be managed by primary anastomosis, much more modest esophageal atresia gaps between 1 cm

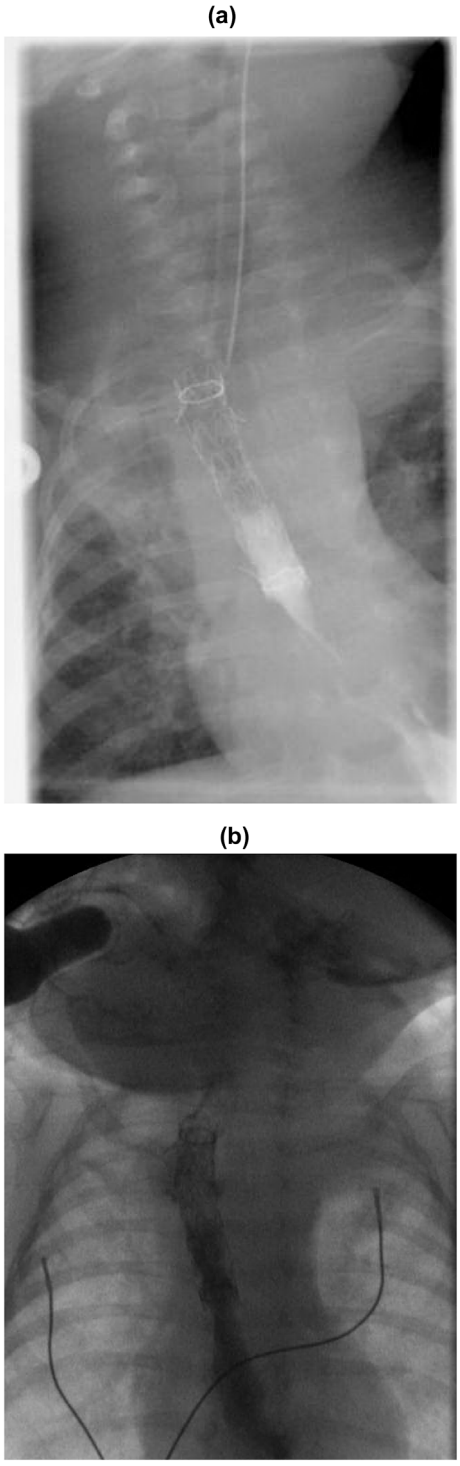

Fig. 2. (a) – During placement of Viabil biliary stent for anastomotic stricture. **(b) –** Repeat imaging four days after stent placement.

and 2 cm likewise experienced postoperative stricture at a rate of 22.5% [12]. Therefore, close follow-up and management of postoperative esophageal stricture is required.

Strictures are initially managed with endoscopic dilation, such as endoscopic balloon dilators or Savary-Gilliard dilators (Cook Medical; Bloomington, IN). Comparisons of balloon dilators and Savary-Gilliard dilators have shown that patents undergoing balloon dilation required fewer procedures compared to patients undergoing bougie dilation [6], and that the balloon dilators demonstrated fewer technical failures