

Çocukluk Çağı Özofagus Darlıklarda Tekrarlanan Savary-Gilliard Buji Dilatasyonları Etkili Midir?

Repeated Bougie Dilation with Savary-Gilliard Dilators in the Treatment of Pediatric Esophageal Strictures: How Effective is it?

Veli Avcı¹, Burcu Güven²

¹Van Yüzüncü Yıl Üniversitesi, Tıp Fakültesi, Çocuk Cerrahisi Ana Bilim Dalı, Van

²Van Yüzüncü Yıl Üniversitesi, Tıp Fakültesi, Çocuk Gastroenteroloji Bilim Dalı, Van

ÖZ

GİRİŞ ve AMAÇ: Çocukluk çağı özofagus darlıkları ile ilgili veriler yetersizdir. Bu çalışmada amaç, çocukluk çağında farklı sebeplerden dolayı oluşan özofagus darlıklarında tekrarlanan Savary-Gilliard buji dilatasyon işlemlerinin etkinliğini saptamaktır.

YÖNTEM ve GEREÇLER: 2012-2018 yılları arasında özofagus darlığı saptanan ve tekrarlanan buji dilatasyon programına alınan hastalar retrospektif olarak incelendi. Hastaların demografik özellikleri, şikayeti, darlığın uzunluğu, etyolojisi, yeri, uygulanan endoskopik dilatörlerin ortalama çapı-uygulama sayısı, uzun dönem kontrol sonuçları ve komplikasyonları değerlendirildi.

BULGULAR: Çalışmaya dahil edilen 25 hastanın 15 (%60)'ü erkek, 10 (%40)'i kız ve ortalama yaş 24 (5-132) ay idi. Hastaların hepsi yutma güçlüğü şikayeti ile başvurular. Etiyolojide en sık (n=18, %72) neden özofagus atrezi cerrahisi komplikasyonuydu. Uygulanan buji dilatörlerin çapı ortalama 11,4±2,32 (5-15) mm olup; her hasta için 2,2 (1-7) defa dilatasyon işlemi gerçekleştirildi. Dar özofagus segmentinin ortalama uzunluğu 15,84±7,97 (5-32) mm idi. Hastaların bir yıllık izlemlerinde 21 (%84) hastada şikayetleri tamamen geçti. Hiçbir hastamızda komplikasyon görülmedi.

TARTIŞMA ve SONUÇ: Özofagus darlığı gelişen çocuk hastalarda endoskopik buji dilatasyonu ideal bir tedavi yöntemidir. Savary-Gilliard dilatörleriyle tekrarlanan buji dilatasyonu basit, pratik, uygun maliyetli ve etkili bir tekniktir.

Anahtar Kelimeler: Özofagus, darlık, dilatasyon, Savary-Gilliard buji, çocuk.

Yayın hakları Güncel Pediatri'ye aittir.

Sorumlu yazar yazışma adresi: Veli AVCI. Van Yüzüncü Yıl Üniversitesi Tıp Fakültesi, Çocuk Cerrahisi AD, Van,

Türkiye

E-posta:

SUMMARY

INTRODUCTION: Data on pediatric esophageal strictures is limited. The aim of this study was to investigate the efficiency of repeated bougie dilation with Savary-Gilliard dilators in the treatment of pediatric esophageal strictures caused by various etiologies

METHODS: The retrospective study included patients who underwent bougie dilation with Savary-Gilliard dilators due to esophageal strictures between January 2012 and July 2018. Demographic and clinical characteristics including age, gender, stricture length, etiology and localization, mean diameter of endoscopic dilators, mean number of endoscopies, and long-term outcomes and complications were reviewed for each patient.

RESULTS: The 25 patients comprised 15 (60%) boys and 10 (40%) girls with a median age of 24 (range, 5-132) months. Dysphagia was the only presenting symptom in all the patients. The most common etiology was primary repair of esophageal atresia (n=18; 72%). Mean dilator diameter was 11,4±2,32 (range, 5-15) mm and the mean number of endoscopies performed was 2,2 (range, 1-7) per patient. The mean length of the narrow esophageal segment was 15,84±7,97 (range, 5-32) mm. The complaints were completely resolved in 21 (84%) patients over a one-year follow-up period. No complication occurred in any patient.

DISCUSSION and CONCLUSION: Endoscopic bougie dilation is an ideal treatment method for pediatric esophageal strictures. Repeated bougie dilation with Savary-Gilliard dilators is a simple, practical, cost-effective, and effective technique.

Keywords: Esophagus, stricture, dilation, Savary-Gilliard bougie, pediatric.

Introduction

Esophageal strictures can have numerous causes including esophageal and gastric surgeries, caustic ingestions, collagen vascular disease, and infectious esophagitis (1). Dysphagia is the most common symptom of esophageal strictures and the diagnosis is primarily established by appropriate imaging studies followed by endoscopy (2,3). Prior to surgical intervention, dilatation is attempted as the initial treatment, which is often performed using bougie dilators at varying sizes depending on the size of the stricture (4,5).

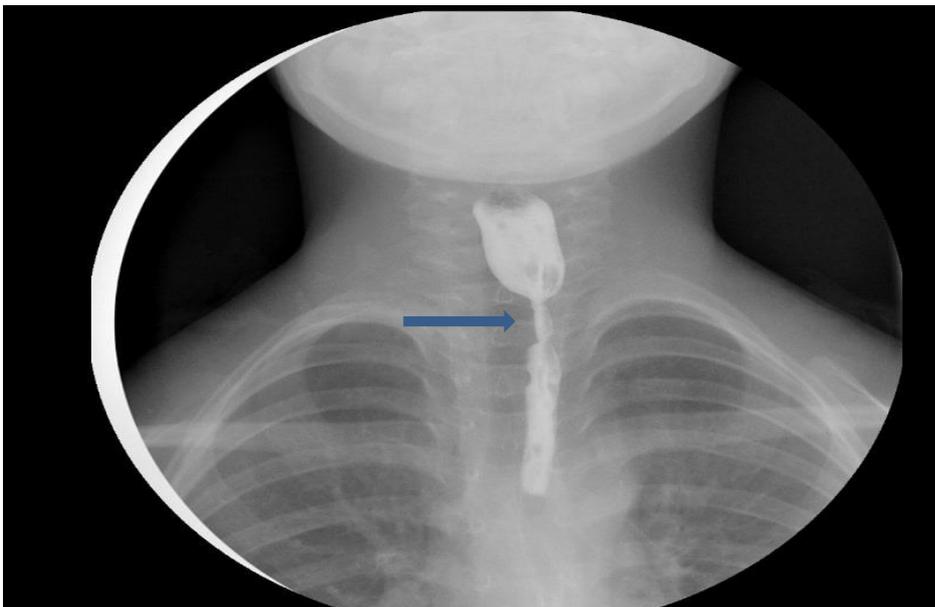
In this study, we aimed to investigate the efficiency of repeated bougie dilatation with Savary-Gilliard dilators in the treatment of pediatric esophageal strictures caused by various etiologies and also to contribute to the literature by presenting our experience in the administration of these dilatations in pediatric esophageal strictures.

Methods

The retrospective study included patients who underwent bougie dilatation with Savary-Gilliard dilators due to esophageal strictures in the Pediatric Gastroenterology and Pediatric Surgery Clinics at Yuzuncu Yil University Medical School between January 2012 and July 2018. Demographic and clinical characteristics including age, gender, stricture etiology and localization, mean diameter of endoscopic dilators, mean number of endoscopies, mean length of stricture and long-term outcomes and complications were reviewed for each patient.

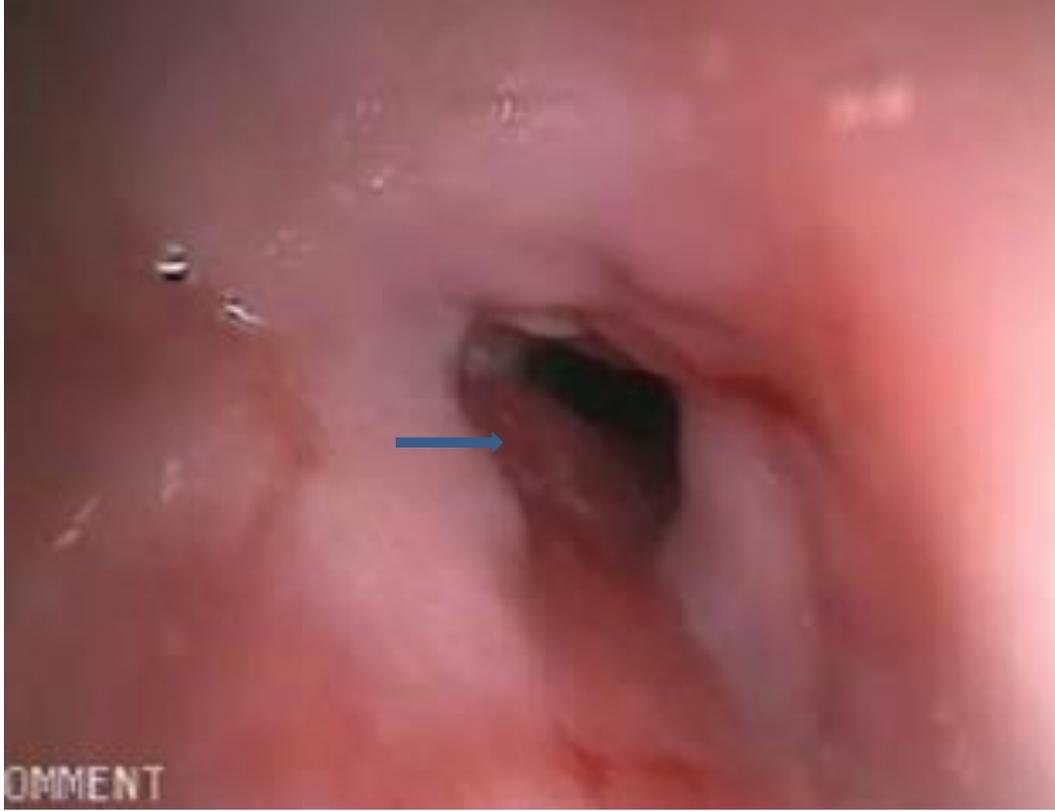
The diagnosis of esophageal strictures was established using esophageal barium radiography (Figure 1).

Figure 1. Barium esophagogram in patient shows a focal stricture



All the sessions were performed under general anesthesia and the stricture orifice was viewed by flexible endoscopy (FUJINON FUJIFILM Corporation 2500) (Figure 2).

Figure 2. Endoscopic view for focal stricture



By laryngoscopic monitoring of the esophagus, a guidewire was passed down to the distal end of the esophagus. The dilatation process was initiated using a Savary-Gilliard bougie dilator lubricated with vaseline, at a smaller size according to the age and body weight of the patient (Figure 3). After each dilatation, the dilator was cleaned with a moist sponge to assess bleeding. The dilator was upsized in consecutive steps and the “rule of three” was applied in each session. Length of stenosis was estimated by endoscopic imaging and radiography. The patient was followed up for 24 h for bleeding control and due to the risk of esophageal perforation and other complications. The sessions were performed according to the severity of dysphagia at four to six week intervals.

Figure 3. The Savary-Gilliard bougie dilators and equipments

The dysphagia score was used for this purpose (Table 1) (6). Early dilatation was performed in patients with a dysphagia score greater than 2. successful dilatation was defined as having no difficulty in swallowing solid food over a period of six months.

Table 1. Dysphagia score

Dysphagia score	Description
0	No dysphagia: able to eat normal diet
1	Moderate passage: able to eat some solid foods
2	Poor passage: able to eat semi-solid foods
3	Very poor passage: able to swallow liquids only
4	No passage: unable to swallow anything

Rule of three: This rule is only applied to wire-guided bougie dilators, not to balloon dilators. An ideal dilatation procedure is performed by starting with a bougie dilator that has nearly the same diameter as the lumen and to continue with progressively larger dilators until resistance to the passage of the dilator is encountered. The dilator that leads to resistance to passage is labeled as the first dilator, and subsequently, second and third dilators of successive size are administered, thereby totaling three dilators. Moreover, care should be taken to avoid skipping a dilator size (7).

Ethics Approval: The study was conducted in accordance with the 1964 Declaration of Helsinki and was approved by the local ethics committee (Ethics approval number 09-11-2018/08). Also in the study, informed consent was obtained from the legal guardians of all individual participants.

Statistical analysis: Data were analyzed using SPSS for Windows version 21,0 (IBM Statistics, Armonk, NY). Continuous variables were expressed as mean, standard deviation (SD), minimum, and maximum. Categorical variables were expressed as frequencies and percentages. Comparison of continuous variables with regard to dilatation success was performed using Student-T test. Relationships between categorical variables were determined using the Chi-square Test. A p-value <0,05 was considered as statistically significant.

Results

The 25 patients comprised 15 (60%) boys and 10 (40%) girls with a median age of 24 (range, 5-132) months. Dysphagia was the only presenting symptom in all the patients. The most common etiology was primary repair of esophageal atresia (n=18; 72%), followed by caustic ingestions (n=5; 20%), and surgery of hiatal hernia (n=2; 8%). In terms of dilatation success, a significant effect was not observed in the patients ($p=0,62$).

Mean dilator diameter was $11,4\pm 2,32$ (range, 5-15) mm and the mean number of endoscopies performed was 2,2 (range, 1-7) per patient. As the dilator diameter and the number of endoscopies increased, the dilatation success also increased ($p=0,01$). However, mean length of stricture was 15.84 ± 7.97 (range, 5-32) mm, and there was a negative correlation between length of stricture and dilatation success ($p=0,54$). The stricture was localized in the cervical esophagus in 21 (84%) and in the thoracic esophagus in 4 (16%) patients.

No pneumothorax or other complications were detected in the anterior-posterior chest radiography obtained after the procedure. In additionally, no complications such as esophageal perforation, chest pain, vomiting or mortality occurred in any patient. The complaints were resolved (dysphagia score: 0-1) in 21 (84%) patients over a one-year follow-up period while the remaining 4 (16%) patients that dysphagia scores ≥ 2 , are still being treated.

Discussion

The first dilatation reported in the literature was performed by pushing “whale bone” down the esophagus in the 17th century by an Italian anatomist known as Fabricius ab Aquapendente, who used a wax bougie as a rigid dilator. The bougienage procedure for the dilatation of esophageal strictures was first published in 1821 (8).

Esophageal stricture can be caused by numerous etiologies including primary repair of esophageal atresia, caustic ingestions, esophageal fibrosis and scarring secondary to gastric surgery (9). In our patients, primary repair of esophageal atresia was the most common etiology. Moreover, the inclusion of the cases caused by caustic ingestions and hiatal hernia surgery was considered to widen the patient spectrum of the study.

Esophageal stricture may result in several complications including dysphagia, food impaction, and chest pain (7). Progressive dysphagia to solid food is the most common cardinal symptom (2,7). In our patients, dysphagia was the only presenting symptom, which could be attributed to the fact that all the patients were follow-up patients and thus the symptoms other than dysphagia were promptly managed before becoming apparent. Moreover, all of our patients were too young to describe symptoms such as odynophagia.

The diagnosis of esophageal strictures is primarily established by appropriate imaging techniques followed by endoscopy (3). Of these techniques, barium esophagography is the primary method of choice which provides information on the number, length and diameter, and localization of lesions at the stricture site and on the presence of other lesions, whether the esophageal wall is regular, and the lumen diameter. The data obtained on esophagography should be merged with endoscopic findings. Endoscopy is often performed prior to dilatation when no additional suspicion is detected on barium esophagography. Endoscopy is typically used for the confirmation of the diagnosis and also for treatment. Moreover, endoscopy is more sensitive than barium esophagography in the detection of mucosal lesions that are likely to be missed (9). In our study, the patients were primarily diagnosed by barium esophagography. However, endoscopy was performed only in the patients whose parents provided a written consent. In two patients, the initial dilatations were performed using endoscopy.

Prior to surgical intervention, dilatation should be attempted as the initial treatment (4). Dilatations often performed using Savary-Gilliard bougie dilators at varying sizes depending on the size of the stricture (5,9). The “rule of three” should be followed in each session to ascertain the safety of dilatation (7). The dilatation sessions can be performed at four to six week intervals (5,10,11). However, there are some other reports suggesting that the sessions should be performed at two to three week intervals (12). In our study, no surgical intervention was performed in any patient both before and after dilatation and the sessions were performed at four to six week intervals, as suggested by the majority of the studies in the literature. Moreover, the “rule of three” was followed for each patient and no additional rigid intervention was performed in any patient.

The goal of dilatation is to achieve an esophageal diameter of >12,8 mm in children aged younger than 5 years and a diameter of >14 mm in older children to allow regular food intake (5). In our patients, the mean dilator size was 11,4 mm in children aged younger than five years and 14,3 mm in older children. Dilatation is commonly performed with balloon and bougie dilators. Although balloon dilators provide only segmental benefit at best, bougie dilators can dilate through the entire length of the esophagus. Moreover, balloon dilators are generally more costly compared to bougie dilators and also lead to

increased risk of radiation exposure (11). In contrast, bougie dilators are cost-effective, reusable, and also provide dilatation to the extent of their diameters by applying force in both the longitudinal and radial planes (13). In our patient, we preferred bougie dilators since we had relatively greater experience in the use of these dilators and because they are less costly compared to balloon dilators. Moreover, it should be recognized that balloon dilators cannot achieve the expansion achieved by bougie dilators even when inflated to their maximum extent (13).

A number of complication can occur during dilatation, including hemorrhage, aspiration, and pneumothorax (4,9). Moreover, perforation at the stricture site is the most common cause of mortality (10). For these reasons, excessive force should be avoided during endoscopy (14). In our patients, the dilator was cleansed with a moist sponge after each dilatation to assess bleeding. When blood was detected on the dilator after removing, the dilatation procedure was terminated for that patient, in order to avoid esophageal perforation and hemorrhage. Furthermore, anterior-posterior chest radiography was performed to achieve prompt diagnosis of pneumothorax. Due to these measures, no complication or mortality occurred in any of our patients.

The present study is limited due to its small patient population and retrospective; however, the sole inclusion of pediatric patients could be considered as a remarkable strength of our study.

In conclusion, endoscopic bougie dilatation is an ideal treatment method for pediatric esophageal strictures. Repeated bougie dilatation with Savary-Gilliard dilators is an effective, simple, practical and cost-effective technique. Moreover, performing this technique over a wire and cleansing the dilator with a moist sponge after each dilatation could be useful in the reduction of complications.

Funding: The authors declared that this study received no financial support

Competing interests: The authors declare that they have no competing interest

References

1. Dzeletovic I, Fleischer DE, Crowell MD, Pannala R, Harris LA, Ramirez FC, and Alexander JA. Self-dilation as a treatment for resistant, benign esophageal strictures. *Dig Dis Sci.* 2013;58(11):3218-23.
2. van Halsema EE, t Hoen CA, de Koning PS, Rosmolen WD, van Hooft JE, Bergman JJ. Self-dilation for therapy-resistant benign esophageal strictures: towards a systematic approach. *Surg Endosc.* 2018;32(7):3200-7.
3. Dzeletovic I, Fleischer DE, Crowell MD, Kim HJ, Harris LA, Burdick GE, and Sharma VK. Self dilation as a treatment for resistant benign esophageal strictures: outcome, technique, and quality of life assessment. *Dig Dis Sci.* 2011;56(2):435-40.
4. Koca T, Dereci S, Akçam M. The Successful Treatment of Congenital Esophageal Stenosis by Endoscopic Dilatation. *J Pediatr Res.* 2016;3(2):126-8.
5. Al-Hussaini A. Savary Dilation Is Safe and Effective Treatment for Esophageal Narrowing Related to Pediatric Eosinophilic Esophagitis. *J Pediatr Gastroenterol Nutr.* 2016;63(5):474-80.
6. Chang CH, Chao HC, Kong MS, Chen SY, Chen CC, Lai MW. Clinical and nutritional outcome of pediatric esophageal stenosis with endoscopic balloon dilatation. *Pediatr Neonatol.* 2018.
7. Baron TH. Management of benign esophageal strictures. *Gastroenterol Hepatol (N Y).* 2011;7(1):46-9.
8. Kochman ML, McClave SA, Boyce HW. The refractory and the recurrent esophageal stricture: a definition. *Gastrointest Endosc.* 2005;62(3):474-5.
9. Bilgin Buyukkarabacak Y, Taslak Sengul A, Pirzirenli MG, Basoglu A. Recurrent dilatation in resistant benign esophageal strictures: timing is significant. *Turk J Med Sci.* 2016;46(1):79-83.
10. Köksoy FN, Gönüllü D. The Benign Strictures of the Esophagus. *JAREM.* 2016;6:1-14.
11. Taylor JS, Danzer E, Berquist WE, Wall JK. Dilation of Esophageal Stricture in a Pediatric Patient Using Functional Lumen Imaging Probe Technology Without the Use of Fluoroscopy. *J Pediatr Gastroenterol Nutr.* 2018;67(2):e20-1.
12. Stenstrom P, Anderberg M, Borjesson A, Arnbjornsson E. Dilations of anastomotic strictures over time after repair of esophageal atresia. *Pediatr Surg Int.* 2017;33(2):191-5.
13. Siersema PD. Treatment options for esophageal strictures. *Nat Clin Pract Gastroenterol Hepatol.* 2008;5(3):142-52.
14. Bakken JC, Wong Kee Song LM, de Groen PC, Baron TH. Use of a fully covered self-expandable metal stent for the treatment of benign esophageal diseases. *Gastrointest Endosc.* 2010;72(4):712-20.