SURGICAL MEDICAL SCIENCES / CERRAHİ TIP BİLİMLERİ

# Endoscopic Balloon Dilation of Esophageal Strictures in Children with Esophageal Atresia: 19 Years' Experience

Özofagus Atrezili Çocuklarda Özofagus Darlıklarında Endoskopik Balon Dilatasyonu: 19 Yıllık Deneyim

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# Abstract

**Objectives:** Endoscopic balloon dilation (EBD) is a frequently used method in the treatment of esophageal strictures in patients with esophageal atresia. The aim of this article is to convey our 19 years of experience in EBD performed in patients with esophageal atresia.

**Materials and Methods:** Thirty-seven patients with esophageal atresia who developed esophageal stricture and underwent EBD between 2003 and 2022 were included in the study. The number of EBD sessions, the time elapsed between dilations, the size of the dilated balloon, the development of complications after the procedure, additional surgical intervention and the follow-up periods were evaluated.

**Results:** In patients with esophageal atresia, EBD was performed at an average of 0.72-month (2 weeks-29 months) intervals, average of 3.3 months after the primary repair. EBD was performed only one time in 6 patients (16.2%), after the single procedure, the patients did not have clinical symptoms or need for dilation. Time between two last dilation procedure is 4.5 months (1-48). Esophageal perforation was seen in 7 cases (8.8%) and was managed with conservative treatment. In 14 of the patients, it was observed that the need for dilation was continued.

**Conclusion:** EBD is minimally invasive and safe method for esophageal strictures after primary esophageal atresia repair. It prolongs the time required for dilation with a higher success rate in anastomotic strictures. The optimal number of dilations should be determined and accompanying comorbidities should be treated primarily.

Key Words: Children, dilation, endoscopy, esophageal atresia, stricture

# Öz

Amaç: Endoskopik balon dilatasyonu (EBD), özofagus atrezili hastalarda özofagus darlıklarının tedavisinde sıklıkla kullanılan bir yöntemdir. Bu makalenin amacı, özofagus atrezili hastalarda uygulanan EBD'deki 19 yıllık deneyimimizi aktarmaktır.

Gereç ve Yöntem: 2003-2022 yılları arasında özofagus darlığı gelişen ve EBD uygulanan 37 özofagus atrezili hasta çalışmaya dahil edildi. EBD seans sayısı, dilatasyonlar arasında geçen süre, dilate balonun boyutu, işlem sonrası komplikasyon gelişimi, ek cerrahi girişim ve takip süreleri değerlendirildi.

**Bulgular:** Özofagus atrezisi olan hastalarda primer onarımdan ortalama 3,3 ay sonra, ortalama 0,72 ay (2 hafta-29 ay) aralıklarla EBD uygulandı. Altı hastada (%16,2) sadece bir kez EBD uygulandı ve bu hastalarda klinik semptom veya dilatasyon ihtiyacı görülmedi. Son iki dilatasyon işlemi arasında geçen süre 4,5 aydı (1-48). Yedi olguda (%8,8) dilatasyon işlemi sonrası özofagus perforasyonu görüldü ve konservatif olarak tedavi edildi. Hastaların 14'ünde dilatasyon ihtiyacının devam ettiği görüldü.

**Sonuç:** EBD, özofagus atrezisi primer onarımı sonrası özofagus darlıkları için minimal invaziv ve güvenli bir yöntemdir. Anastomoz darlıklarında daha yüksek başarı oranı ile dilatasyonlar arası geçen süreyi uzatır. Optimal dilatasyon sayısı belirlenmeli ve öncelikle eşlik eden komorbiditeler tedavi edilmelidir.

Anahtar Kelimeler: Çocuklar, dilatasyon, endoskopi, özofagus atrezisi, darlık





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# Introduction

Esophageal strictures are seen due to different congenital and acquired etiologies in childhood and adolescence. Esophageal strictures, especially strictures developing in the anastomotic line, are the main complications after surgical repair of esophageal atresia.

It has been shown that esophageal stricture occurs in 18% to 50% of patients undergoing esophageal atresia repair (1).

The survival rate of infants born with EA has improved significantly, from 80% to over 95%, in recent years, despite this high rate of complication, with recent advances in surgical techniques and neonatal care (2).

In the treatment of strictures, open surgical methods, dilatation with bougie, dilatation with balloon catheter and fluoroscopic dilations are applied. It is accepted that minimally invasive surgery should be preferred more frequently, especially in pediatric patients. As a result of this approach, it has made open surgery much less preferable by highlighting bouginage and endoscopic dilatation in the treatment of esophageal strictures (3). Since the first successful Endoscopic balloon dilation (EBD) in children, many studies have compared bouginage and EBD in the treatment of esophageal strictures.

Bougie dilation was the first procedure widely available to dilate esophageal strictures. Since the first successful endoscopic dilation in the treatment of esophageal strictures, many studies have evaluated the results of EBD as an alternative to bouginage (4-6). In some studies comparing the two methods, it has been reported that, although there are no significant differences, EBD has a higher success rate and a lower complication rate and reduces the number of dilations (7-9). Fluoroscopy-guided EBDs under general anesthesia are becoming a more preferred method today (8,9).

The risk of complications such as perforations and the need for fluoroscopic balloon dilation increases in recurrent strictures. Complications caused by frequent exposure to anesthetic substances and psychological problems in children who have to enter the operating room frequently can also increase. This leads us to the use of agents such as steroids and mitomycin C which have been used to inhibit new collagen and stricture formation as conservative treatment. Recurrent strictures are also common in patients with gastroesophageal reflux disease (GERD) due to increased acid exposure. This shows the necessity of antireflux treatment in patients with esophageal atresia and GERD (10).

Strictures are performed at different frequencies and balloon sizes according to the need determined by the length, location, comorbidities differences also change the frequency of dilation, complications and approaches to these complications. We have previously reported our clinical experience to demonstrate the efficacy and safety of fluoroscopic balloon dilation by evaluating the outcomes of patients undergoing fluoroscopic balloon dilation due to anastomotic stricture or corrosive ingestion (11).

The aim of this article is to present the experience of our clinic in fluoroscopic balloon dilation, which is accepted as a current and safe approach in the treatment of esophageal strictures with esophageal atresia, by presenting the results of 19 years.

# **Materials and Methods**

We retrospectively reviewed the medical records of 37 patients with esophageal atresia who underwent fluoroscopic balloon dilation at our institution between 2003 and 2022. The need for dilation of patients with suspected esophageal stricture was determined according to symptoms such as dysphagia score (simplified dysphagia severity rating scale), decreased tolerance depending on the consistency and caliber of the food pieces, and water soluble upper gastrointestinal (GI) contrast radiographies were deployed each esophageal atresia patient one week after primary repair, then when patients are symptomatic.

All of the patients' demographic features, primary diagnosis, additional diseases, level of stricture, total number of dilations, balloon sizes used in the first and last dilation, time between dilations, complications due to the procedure, gastrostomy opening, need for additional surgical procedure, therapeutic agent injection, trial of C-mac, preoperative upper GI contrast radiographies, time spent without the need for dilation, and the follow-up time were reported.

All the esophageal dilation procedures were performed by using upper GI flexible endoscopy with endotracheal intubation under general anesthesia by experienced surgical and anesthesia team. The informed consent form was obtained from the firstdegree relatives of the patients.

The flexible endoscope (Olympus, Tokyo, Japan) is advanced through the mouthpiece and the balloon catheter (Boston Scientific, Cork, Ireland) was passed through the working channel of the endoscope. Then the balloon catheter was advanced through the stricture and balloon was inflated with radiopaque contrast solution. Before inflation, correct placement of the deflated balloon was checked fluoroscopically using an intraoperative mobile C-arm fluoroscopy machine which is compatible with the operating table. The length of the balloon was 5.5 cm, and the diameters range from 6 to 20 mm and were translucent in color. The balloon was inflated up to 4-10 atm pressure and kept inflated for 5 min. After confirming the persistence of the "waist" and it was both endoscopically and fluoroscopically safe, the diameter of the balloon was increased. Appropriate balloon catheter size was selected by the surgeon by evaluating the patient's age, body weight, the severity of the esophageal stricture determined on the fluoroscopic image, and the balloon catheter sizes used in the patient's previous dilations. Usually the procedure lasted 15 to 20 minutes.

Antibiotic prophylaxis was not routinely given unless there were any complications. All patients underwent chest X-ray 2 hours after the procedure. If no pathological finding is detected in the patients examination and chest X-ray, the patients were started to feed orally with soft food and discharged 3-4 hours after the procedure. Upper Gl contrast study was not performed routinely in asymptomatic patients. Patients with severe strictures were called back after 4-6 weeks. Symptom monitoring was recommended for patients with mild strictures and no need for dilation for more than 3 months.

#### Results

Total of 37 patients who underwent fluoroscopy-assisted endoscopy dilation due to esophageal stricture between 2003 and 2022 were included in the study. The patients underwent fluoroscopy-assisted endoscopy dilation an average of 6 times per patient, with an average of 0.72 months between the first dilation and the last dilation procedure (Table 1).

In patients with esophageal atresia, dilation was performed at an average of 0.2 month (2 weeks-29 months) intervals, average of 3.3 months after the primary repair. Endoscopic dilation was performed only one time in 6 patients (16.2%), after the single procedure, the patients did not have clinical symptoms or need for dilation. Patients were dilated with an average of 6-8 mm balloon catheter in the first procedure, and an average of 12-15 mm balloon catheter in the last procedure (Table 2). In 31 patients, larger size balloon catheter was used in repetitive procedures. Balloon dimensions could not be standardized because they differ according to patients age, degree of stricture.

No complication was seen in 86% of the patients after the procedures (n=32). Esophageal perforation after balloon dilation was seen in 4 cases (10.8%), anastomotic leakage in one case (2.7%). No additional surgery was performed after endoscopic dilation in any of the patients with esophageal atresia and all the complications managed with conservative treatment. Oral nutrition was stopped, parenteral nutrition was started, and broad-spectrum antibiotics were started after consultation with pediatric infectious diseases.

Segmental resection and primary anastomosis was performed in 2 patients (5.4%) and colonic interposition in 2 patients (5.4%) who did not benefit from endoscopic dilation. Gastric pull-up was performed in 2 patients (5.4%) with isolated esophageal atresia. The patients received antireflux treatment in the first year of their lives. Despite this, fundoplication surgery was performed on patients who developed GERD. Nissen fundoplication was performed in 4 patients with severe GERD. An average of 5 dilatations were performed in 4 patients with accompanying GER, and it was observed that stricture also developed in the distal esophagus in these patients. Gastrostomy tube was totally inserted in two patients (5.4%) who could not be fed orally and had failed endoscopic dilation. Then, the gastrostomy tubes of the patients who did not need it were removed. Except for 4 patients, they are fed orally.

| Table 1: Patients, location of stricture, number of dilatations, balloon size, follow-up |                       |                          |  |                        |           |                               |  |  |  |
|--|-----------------------|--------------------------|--|------------------------|-----------|-------------------------------|--|--|--|
| Pathology  | Esophageal<br>atresia | Epidermolysis<br>bullosa | Caustic strictures                                     | Cricopharyngeal<br>bar | Achalasia | Aberrant<br>subclavian artery |  |  |  |
| Patients (n)   | 37                    | 21                       | 14   | 4                      | 2         | 1                             |  |  |  |
| Sex (F/M)  | 16/21                 | 10/11                    | 8/6  | 3/1                    | 0/2       | 1/0                           |  |  |  |
| Location of the stricture (cm)   | 13.6*                 | 15.1                     | 21   | 10.3                   | 32        | 15                            |  |  |  |
| Number of dilatation   | 6                     | 5                        | 18   | 4.7                    | 1.5       | 7                             |  |  |  |
| Balloon size (first)   | 6-8                   | 10-12                    | 10-12  | 8-10                   | 15-18     | 8-10                          |  |  |  |
| Balloon size (last)  | 12-15                 | 15-18                    | 18-20  | 12-15                  | 18-20     | 18-20                         |  |  |  |
| Time between first and last dilatation (month)   | 0.72                  | 3.8                      | 2.3  | 1.6                    | 0.3       | 2.2                           |  |  |  |
| Time between the last two dilatations (month)  | 2.9                   | 6.5                      | 5  | 5                      | 6.5       | 8                             |  |  |  |
| Intralesional injection  | 5<br>(steroid)        | -                        | 11 (steroid (n=6);<br>steroid and<br>mitomycin C (n=5) | -                      | -         | -                             |  |  |  |
| Follow-up (months)   | 43.6                  | 59.2                     | 52.2   | 35                     | 53        | 29                            |  |  |  |
| *: - Usually, the anastomosis, F/M: Female/male  |                       |                          |  |                        |           |                               |  |  |  |

| Table 2: Complications, success, additional surgery |  |                          |  |                        |             |                                  |  |  |  |
|---|--|--------------------------|--|------------------------|-------------|----------------------------------|--|--|--|
| Orally fed<br>patient                               | 33<br>(89.1%)  | 18<br>(85.7%)            | 14<br>(100%)   | 3<br>(75%)             | 2<br>(100%) | 1<br>(100%)                      |  |  |  |
| Additional<br>surgery                               | Reanastomosis - 2;<br>colonic interposition - 2;<br>gastric pull-up - 2;<br>Gastrostomy - 10 | Gastrostomy - 4          | Gastrostomy<br>- 3;<br>colocin<br>interposition<br>- 1;<br>stent - 1 | Gastrostomy - 1        | -           | -                                |  |  |  |
| Success rate, n<br>(%)                              | 36<br>(92.3%)  | 13<br>(61.9%)            | 10<br>(71.4%)  | 3<br>(75%)             | 2 (100%)    | 1 (100%)                         |  |  |  |
| Complications/<br>treatment                         | 5 (perforation/<br>conservative)   | -                        | 2 (perforation/<br>conservative)                                     | -                      | -           | -                                |  |  |  |
| Patients  | Esophageal atresia   | Epidermolysis<br>bullosa | Caustic<br>strictures  | Cricopharyngeal<br>bar | Achalasia   | Aberrant<br>subclavian<br>artery |  |  |  |

Intralesional steroid injection was applied to the stricture area in the following fluoroscopic dilation procedures in 5 patients who developed strictures in the early period and needed frequent dilatation. The injection was applied to 2 patients with the first endoscopic dilation procedure, and 3 patients after an average of 3.8 (1-12) endoscopic dilation sessions. After the injections, an average of 1.2 (2-3) endoscopic dilations were performed. Triamcinolone acetonide (Sinecord<sup>®</sup>) was used for intralesional steroid injection at a dose of 0.5 mg/kg (total dose of 40 mg), divided into four equal amounts and injected in four quadrants of the esophagus at the proximal edges of the stricture before dilatation and the total number of injection sessions differed individually according to dysphagia relief.

The dilation need of the patients was determined primarily according to the clinical symptoms. Upper GI contrast study was performed in all patients.

In the follow-ups, in one patient (2.7%), a total of 8 dilatation procedures were performed from January 2020 to August 2022. The clinical symptoms of the patient were evaluated and it was observed that the need for dilatation still continued. One patient with VACTERL association and one patient with Fallot Tetralogy died due to comorbidities.

# Discussion

Today, the development of minimally invasive surgery shows its effects in many areas. Major surgeries performed in the past are less preferred now. Treatment of resistant esophageal strictures was esophageal replacement (6,13,14). In this process, the colon is generally preferred (6,15). The size of the operation, the presence of several anastomoses in different places, the late start of feeding; It creates both intraoperative and postoperative difficulties for these patients who already have malnutrition and generally developmental delay. One of the advantages of the procedure is that oral feeding is possible after two hours (16). In addition, it is more preferred especially for pediatric patients in terms of protecting the physiology and anatomy of another part of the intestinal system (3).

It is seen that intraoperative and postoperative complications in EBD are lower than other methods (3,15,17). The question of whether the dilatation procedure should be performed routinely or in symptomatic patients was evaluated by comparing the approaches of different centers from the study of Koivusalo et al. (18), and it was concluded that dilation would be the right approach in case of symptom development.

This situation indirectly affects the length of hospital stay (8,19). Since the esophagus could not be visualized in dilation with bougie, it was observed that the perforation was more (14,17,19,20). In esophageal replacement, anastomotic leakage, anastomotic stricture, mediastinitis, ileus etc. Complications such as these are seen, medical and surgical methods are preferred (3,15). Simultaneous endoscopic and fluoroscopic imaging of the esophagus in EBD reduces the complication rate.

When we look at the literature, the success rate of EBD is 92-100%; dilatation with bougie 87%; esophageal replacement appears to be 89% (4,19,21-23). In our study, 30 patients discontinued the follow-up due to the lack of clinical problems. The acceleration of anabolic processes, thanks to the sufficient calories they take in patients whose swallowing problem improves, causes weight gain, improvement of growth retardation and indirectly an increase in the quality of life (16).

When we look at current research, although there are many large series of publications, there are important topics of discussion. Inflating time of the balloon, reprocessing interval, necessity of contrast radiography before the procedure etc. Although new studies suggest that the balloon should be followed with swelling in the mean of 15 minutes, we think that earlier deflating at the upper end of the esophagus would be beneficial. This is important in order to avoid possible complications by preventing vagal compression. With endoscopic and fluoroscopic imaging, the inflation rate of the balloon can be easily evaluated and decided. We think that this is one of the reasons that reduces the complication compared to dilation with bougie.

In the literature, it was observed that the recovery period after the development of damage to the esophageal lumen is 6-24 months (24). Recurrence of stricture can be prevented by endoscopic dilations performed at regular intervals during the remodeling process (25). In another literature, it was reported that it was necessary to wait 2 years before deciding that endoscopic dilation had failed (26). Esophageal replacement can be performed if it is seen that the esophageal stricture continues and the time between dilation procedures is not shortened although endoscopic dilation procedures are performed at regular intervals for 2 years.

In the study of Pearson et al. (27), it was observed that anastomotic leakage and stricture, which is more common in the lower esophagus, developed in patients with GER disease. This shows that it is necessary to perform antireflux surgery in reflux patients who do not respond positively to medical treatment in order to reduce the need for dilation (27).

In recent years, it has been shown that intralesional or topical drug (such as steroids) applications to the stricture area reduce the number of dilations, prolong the time between dilations and reduce the rate of application for esophageal replacement. (14,28-31). For the first time in 1995, steroid injection was shown to reduce stricture (30). Although a statistical comparison could not be made, the data obtained in this study are similar to other studies and support that early steroid use is more successful.

#### Conclusion

Esophageal strictures are mostly seen at the anastomotic line in patients with esophageal atresia. The dilation needs of patients and the location of esophageal strictures depends on the time of primary esophageal atresia repair, the type of esophageal atresia, and the experience of the operating surgeon. Although EBD is considered as the safest approach in the treatment of strictures, repetitive procedures increase the complication rate. The optimal number of dilations should be determined, accompanying comorbidities should be treated primarily and the treatment success of experienced clinics should be evaluated.

#### Ethics

**Ethics Committee Approval:** The data of the study were retrieved from electronic patient record of hospital authority.

**Informed Consent:** Patient consent was waived by the Ethics Committee.

#### **Authorship Contributions**

Surgical and Medical Practices: U.A., E.E., M.B.K., A.Y., A.M.Ç., G.G., Concept: U.A., E.E., E.Ek., P.K., M.B.K., A.Y., A.M.Ç., G.G., Design: U.A., E.E., E.Ek., P.K., A.M.Ç., G.G., Data Collection or Processing: U.A., E.E., Analysis or Interpretation: U.A., E.E., E.Ek., P.K., M.B.K., A.Y., A.M.Ç., G.G., Literature Search: E.Ek., P.K., Writing: U.A., E.E., E.Ek., P.K.

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