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Our approach to esophageal perforation secondary to dilatation of caustic esophageal stricture in children

Çocuklarda korozif özofajit darlığının dilatasyonuna bağlı gelişen özofagus perforasyonuna yaklaşımımız

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ABSTRACT

Background: This study aims to review our 20-year experience in children with esophageal perforation and develop an algorithm.

Methods: The study included 50 patients (32 boys, 18 girls; mean age 4.7±2.6 years; range 1 to 17 years) with esophageal perforation secondary to dilatation of caustic esophageal stricture which was performed between January 1985 and December 2014 in our department. Patients were evaluated retrospectively according to age, sex, time elapsed from intake of caustic substance until admission, time elapsed from dilatation until the diagnosis of perforation, clinical findings, the location of perforation, and method of treatment.

Results: Diagnosis of perforation was confirmed within 24 hours after dilatation in 40 patients and 24 hours after dilatation in 10 patients. The mortality rate was higher in the late diagnosed group (n=2). Perforations occurred in cervical esophagus in two patients, abdominal esophagus in four patients, and thoracic esophagus in 44 patients. In 21 patients, esophageal perforation healed conservatively and no surgical intervention was required. Of the 29 patients with chest tube, 15 healed with conservative management, while nine were performed thoracotomy with abscess drainage and decortications, and five were performed esophagostomy and gastrostomy. Of the five patients who were performed esophagostomy and gastrostomy, two underwent colon interposition operation and three underwent delayed anastomosis.

Conclusion: Esophageal perforation induced by dilatation of caustic esophageal strictures is a serious problem which has to be promptly diagnosed, individualizing the therapeutic approach according to the condition of each patient.

Keywords: Children; esophageal dilatation; esophageal perforation.

ÖZ

Amaç: Bu çalışmada özofagus perforasyonu olan çocuklarda 20 yıllık deneyimimiz incelendi ve bir algoritma geliştirildi.

Çalışma planı: Çalışmaya korozif özofajit darlığı nedeni ile Ocak 1985 - Aralık 2014 tarihleri arasında kliniğimizde uygulanan dilatasyona bağlı özofagus perforasyonu gelişen 50 hasta (32 erkek, 18 kız; ort. yaş 4.7±2.6 yıl; dağılım 1-17 yıl) dahil edildi. Hastalar yaş, cinsiyet, korozif madde alımından başvuruya kadar geçen süre, dilatasyon işlemi sonrasında perforasyonun tanı almasına kadar geçen süre, klinik bulgular, perforasyonun yeri ve tedavi yöntemi açısından retrospektif olarak değerlendirildi.

Bulgular: Perforasyon tanısı 40 hastada dilatasyondan sonraki 24 saat içinde, 10 hastada dilatasyondan 24 saat sonra konuldu. Daha geç tanı konulan hastalarda mortalite oranı daha yüksek idi (n=2). Perforasyonlar iki hastada servikal, dört hastada abdominal, 44 hastada torasik özofagusta idi. Yirmi bir hastada özofagus perforasyonu konservatif şekilde düzeldi ve cerrahi girişime gerek olmadı. Göğüs tüpü olan 29 hastadan 15'i konservatif tedavi ile düzelir iken dokuz hastaya torakotomi ile apse drenajı ve dekortikasyon, beş hastaya da özofagostomi ve gastrostomi uygulandı. Özofagostomi ve gastrostomi uygulanan beş hastadan ikisine kolon interpozisyon ameliyatı, üçüne gecikmiş anastomoz yapıldı.

Sonuç: Korozif özofajit darlığının dilatasyonuna bağlı özofagus perforasyonu hızlıca tanı konulması gereken ciddi bir sorundur ve tedavi yaklaşımı her hastanın durumuna göre bireyselleştirilmelidir.

Anahtar sözcükler: Çocuklar; özofagus dilatasyonu; özofagus perforasyonu.

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Esophageal perforation is a rare and life-threatening condition. Its incidence is increasing due to the increase in the number of endoscopic procedures.[1] The main causes of esophageal perforation include diagnostic or therapeutic endoscopy or dilatation for caustic injury. Presenting symptoms differ based on localization of perforation and time elapsed from the occurrence of perforation until diagnosis. Rarity of the condition and non-specific symptoms such as fever and pain result in a delay of diagnosis which causes poor outcomes. Despite modern diagnostic methods, the condition still has high mortality rates (19.7%).[2]

Abdominal or thoracic pain, fever, dysphagia, dyspnea, vomiting, and subcutaneous emphysema are the most common symptoms.[3] The initial symptoms resemble postoperative pain or pneumonia after dilatations, which emphasize the importance of considering the risk of perforation after every dilatation. Localization of perforation may also cause a delay in diagnosis.

Early diagnosis and effective treatment are important predictors for survival. Thus, in this study, we aimed to review our 20-year experience in children with esophageal perforation and develop an algorithm.

PATIENTS AND METHODS

Of a total of 275 patients (180 males, 95 females; mean age 4.7±2.6 years; range 1 to 17 years) who underwent dilatation procedures (a total of 2,180 dilatations) in Istanbul University, Istanbul Medical Faculty, Department of Pediatric Surgery between January 1985 and December 2014, perforation occurred in 50 patients (32 males, 18 females; mean age 4.7±2.6 years; range 1 to 17 years) who were retrospectively evaluated according to sex, age, time elapsed from intake of caustic substance until admission, time elapsed from the occurrence of perforation until diagnosis, clinical findings, localization of perforation, and methods of treatment. In the past, for caustic esophageal injuries, we used to evaluate the esophagus within 24 hours with early endoscopy. In grade 2b and 3 injuries, children were being fed with nasogastric feeding tubes for one week, than the tube was removed and oral feeding was allowed one week after the collagen production began to decrease.[4] However, three years ago we changed our policy, giving up early endoscopy and starting feeding earlier as soon as the child can tolerate oral intake. At the end of third week, contrast esophagography is obtained and in case of stricture, long-term dilatation is applied. Dilatation program starts three weeks after the caustic injury and is applied once in three weeks for the first three months. Later on, the intervals between the dilatations are extended and completed within two years.

In this study, the dilatations were performed with antegrade or retrograde bougie or balloon dilators under general anesthesia. Patients with an esophageal perforation owing to dilatation, with intractable strictures or with an irregular shaped esophagus underwent gastrostomy and the dilatations were performed in a retrograde fashion with Tucker® dilators, which carry a lower perforation risk. If available, balloon dilators were used for strictures with low diameters, which were potentially susceptible to perforation. In the remaining patients, antegrade dilator was managed with rush dilators. Antegrade dilatation is routinely performed by wire guidance with Savary® dilators.

After a dilatation procedure; fever, dyspnea, tachycardia, restlessness, or subcutaneous emphysema suggest esophagus perforation. In such patients, a chest X-ray is performed. In case of subcutaneous emphysema, mediastinal enlargement, pericardial air, pneumothorax or pleural effusion, the diagnosis of esophagus perforation is confirmed (Figure 1-3). We did not evaluate contrast esophagography, endoscopy, and contrast computed tomography routinely. Patients diagnosed within 48 hours were considered as early diagnosed, while patients diagnosed after 48 hours were considered late diagnosed. Those patients with suspicion of perforation were fed parenterally with intravenous fluids, parenteral antibiotics, and H2 receptor antagonists. Our aim was to employ a conservative approach to all of our patients. We planned an additional intervention according to the complication; i.e. we placed a chest tube in case of pneumothorax, or a mediastinal tube in case of mediastinal air.

In patients with pachypleuritis or abscess despite drainage, thoracotomy and decortications are applied. When clinical symptoms disappear and drainage from the tube stops, contrast esophagography is evaluated, closure of the perforation is confirmed, and oral feeding is started.

Statistical analysis

Descriptive statistical methods were used. ANOVA and t-tests were used to compare the group distributions and Kolmogorov-Smirnov and Shapiro-Wilk normality tests were used for the normality analyses. The Kruskal-Wallis test was used for the variance analysis and the Mann-Whitney U test was used to compare those groups for which a normal distribution did not exist.

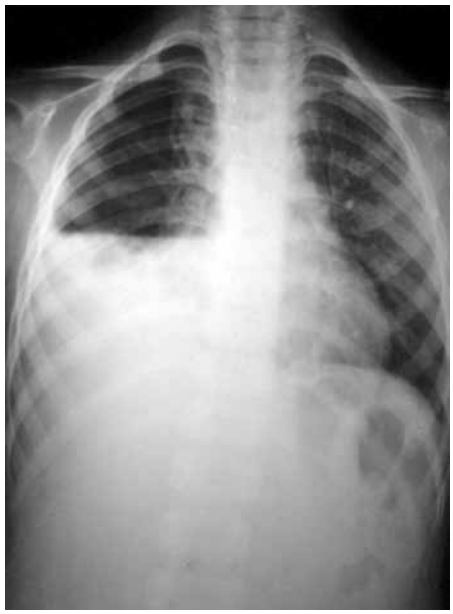

Figure 1. Pleural effusion secondary to thoracic esophageal perforation.

Figure 2. Pneumomediastinitis after thoracic esophageal perforation.

Ninety-five percent was accepted as the confidence interval and a p value ≤0.05 was considered significant for the analysis.

RESULTS

In 71% of children, the caustic material was sodium hydroxide. Other common caustic alkaline substances were potassium hydroxide and sodium bicarbonate, and acidic substances were hydrochloric acid, nitric acid, and sulfuric acid. The length of strictures varied from 3 to 5 cm.

Two hundred and twenty-seven of the 275 patients admitted to our center immediately after caustic ingestion (within 48 hours) and the dilatation was started on the third week promptly. Forty-eight of the 275 patients were referred to us with the diagnosis

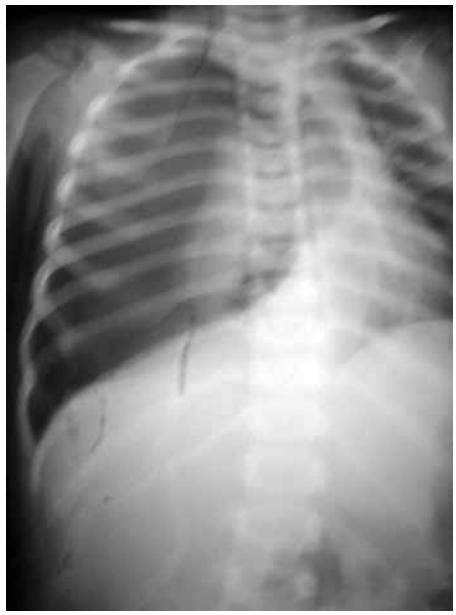

Figure 3. Pneumothorax after perforation.

Figure 4. Stricture after perforation.

Table 1. Perforation rate in late admission group was significantly higher (p<0.0001)

	Late admission	Rate of perforation (Late admission)	Early admission	Rate of perforation (Early admission)	*Total*
	n	%	n	%	n
Presence of perforation	24	50	26	11.45	50
No perforation	24		201		225

of stricture six to 12 weeks after injection and the dilatation was started afterwards. The perforation rates in the early admission and late admission groups were 26/227 (11.45%) and 24/48 (50%), respectively (Table 1). The perforation rate in the late admission group was significantly higher (p<0.0001).

Pain was the most common symptom, presenting in 33 of the 50 patients (66%) with perforation. Other symptoms were pneumoderma in 32 patients (64%), dyspnea in 21 patients (42%), fever in 18 patients (36%), and dysphasia in 13 patients (26%).

Of the 50 patients, perforation was confirmed within 24 hours in 40 (early diagnosis group), while after 24 hours in 10 (late diagnosis group). The mortality rate was 20% (n=2) in the late diagnosed patients which was significantly higher than the early diagnosis group (Table 2). In these two patients, we were able to establish the final diagnosis after 24 hours due to late onset of thoracic symptoms. Both patients had perforation on the thoracic site. While one received chest tube drainage, esophagostomy and gastrostomy, and decortication, the other was only administered chest tube. The reasons of mortality in these two patients were mediastinitis and sepsis. All perforations were located on the stricture site. The locations of perforations are shown in Table 3.

Among 50 patients, 21 with early diagnosis underwent nonoperative treatment. In 29 patients with pleural fluid collection, chest tube drainage was performed. Of these patients, 15 recovered only with chest tube drainage (four received mediastinal drainage at the same time) and needed no further surgical procedure, nine received decortication afterwards (while mediastinal drainage was performed in two, pericardiocenthesis was performed in one at the same time), and five underwent esophagostomy and gastrostomy. Two patients who underwent esophagostomy and gastrostomy subsequently required colon interposition. In those patients, we did not perform esophagectomy and we preserved the native esophagus. Three of them received anastomosis later on. We had no primary anastomosis. Late anastomosis was performed in three patients, all of whom had esophagostomy and gastrostomy prior to surgery. These anastomoses were performed with one-layered separated 4/0 Vicryl sutures. We used no tissue coverage. The numbers of patients who received conservative or operative treatments are listed in Table 4.

Table 2. Mortality in patients

Outcome	Early (n=40)	Late (n=10)	*p**
	n	n	
Mortality	0	2	0.04

* Mortality was significantly higher in patients whose perforations were diagnosed after 24 hours (p<0.05).

DISCUSSION

Esophageal perforation is relatively rare in children than it is in adults. However, corrosive material ingestion among children is still a common and serious problem in developing countries.[5] Esophageal stricture after corrosive ingestion results in increased endoscopic manipulations like long-term dilatations and complications including perforation.[6] The treatment of perforation still remains controversial.[7] Conservative treatments are suggested in children with perforation.[8] Esophageal perforation is a serious condition that needs early diagnosis and management because of its high morbidity and mortality.[9] The first step in the diagnosis of esophageal perforation is suspicion. In case of clinical suspicion, the oral intake of the patient should be prevented. Pain, fever, and pneumoderma are the main symptoms of esophageal perforation. Pneumoderma and local inflammation on the neck occur almost in all cervical perforations. Thoracic pain, back pain, dysphagia and symptoms due to pneumothorax are the common symptoms in

Table 3. Locations of perforation

	Cervical	Thoracic	Abdominal
Number of patients	2	44	4

Table 4. Summary of treatment for esophageal perforation secondary to dilatation for caustic esophageal injuries

Treatment	Surgical intervention	Patients (n=50)
Non-surgical (n=21)	Follow-up	21
Surgical (n=29)	Only chest tube	11
	CTD + mediastinal drainage	4
	CTD + decortication	6
	CTD + decortication + mediastinal drainage	2
	CTD + decortication + mediastinal drainage + pericardiosynthesis	1
	CTD + esophagostomy + gastrostomy colon interposition	2
	CTD + esophagostomy + gastrostomy anastomosis	3

CTD: Chest tube drainage.

thoracic perforations. Acute abdominal symptoms are common in abdominal perforations. Early diagnosis is possible with early physical examination and radiological interventions.

For the past three years, all dilatation procedures included either bougie or balloon usage under guidance of a previously placed wire through the stricture. We believe that thanks to this method, we have not experienced any perforation episode since. Likewise, Bicakci et al.[5] reported no perforation in their large series of esophageal balloon dilatation.

Perforation due to instrumentation may occur from the most weak or the pathological sections of the esophagus usually from the cervical or the thoracic region. In rupture due to disintegrity of the esophageal wall, the gastrointestinal content leaks to the body. Initially; chemical mediastinitis, pneumoderma, and peritonitis may develop. Afterwards, with the spread of the aerobic and anaerobic infection, sepsis and shock may occur.

The treatment of esophageal perforation varies according to the length and site of perforation, and degree of contamination; therefore, the treatment is individualized.[10] We used different approaches for each of our patients starting from the most conservative treatment for minimal leakages and proceeding to invasive methods for massive leakages.

Time elapsed from caustic ingestion until the first dilatation is also an important prognostic factor. Forty-eight of the 275 patients were referred to us with the diagnosis of stricture six to 12 weeks after ingestion of caustic substance and the dilatation was started afterwards. In our study, the perforation rate was significantly higher in the late admission group and earlier studies by Gün et al.[4] support our data.

In our series, thoracic region was the most common site of perforation. Panieri et al.[11] reported eleven children in two of which the cervical esophagus and in nine of which the thoracic esophagus were involved. The most common symptom in our study was thoracic pain, followed by fever, dysphagia, and dyspnea.

For diagnosis, the presentation of symptoms after instrumentation and the suspicion of perforation are necessary. As Gander et al.[12] stated, if a patient is complaining of chest pain after upper endoscopy, he/she has an esophageal perforation until proven otherwise. Chest X-ray should be carried out in such cases. We did not perform esophagography routinely to avoid contamination.

Conservative treatment consists of cessation of oral intake, use of antibiotics, and total parenteral nutrition. We preferred conservative treatment in patients suspected of perforation or in cases with minimal symptoms. All of the nonoperatively managed patients were early diagnosed. Amudhan et al.[13] reported a rate of 35% for nonoperative treatment in their series. In accordance with the literature, 42% of the patients were treated nonoperatively in our series.

Two patients (4%) died after perforation due to complication of mediastinitis. Thus, early diagnosis of perforation is an important factor in the outcome. Mortality was significantly higher in patients whose perforation was diagnosed after 24 hours. Similarly, Vieira et al.[14] have shown that delay of diagnosis of perforation is associated with mortality.

In both of the two patients who died in our series, perforations were on thoracic site, they were late diagnosed, and their mortality reason was septic shock following mediastinitis. We believe that late diagnosis increased mortality due to the contamination of the mediastinum after perforation.

As Elicevik et al.[15] reported in their series of 22 children with esophageal perforation, perforation

of the esophagus during the first dilatation session is the most common. All of the children required ongoing treatment for esophageal stricture. Since there is always a risk of second perforation in patients who undergo gastrostomy, a nasal guide is placed for retrograde dilatation.

Conservative treatments are advised in children whereas primary repair is the first choice in adults.[16] Except for two of our patients who underwent colon interposition, all perforations recovered with selective drainage procedures. Therefore, we suggest that following and applying the appropriate drainage procedure should be the first choice.

In conclusion, balloon dilatation or bougienage with guide wire should be used in esophageal dilatations since they have almost zero perforation rates. When the dilatation program is started earlier, the outcome might favorable with lower perforation rates within three weeks. Early diagnosis of perforation secondary to caustic esophageal injury is important for preventing morbidity and mortality since delay of diagnosis is associated with mortality. Moreover, children with esophageal perforation should be managed individually according to the time of diagnosis, length and site of perforation. Resection and anastomosis or transposition may be preferred for perforations in which esophageal continuity is disturbed, otherwise drainage methods and conservative management should be the first choice.

Declaration of conflicting interests

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