Surgical Endoscopy (2023) 37:9291–9298
https://doi.org/10.1007/s00464-023-10489-w

Outcomes of pneumatic dilation in pediatric caustic esophageal strictures: a descriptive and analytic study from a developing country

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Received: 22 May 2023 / Accepted: 23 September 2023 / Published online: 26 October 2023

Abstract

Objectives Corrosive substance ingestion in children represents a significant public health issue due to its long-term health sequelae. Esophageal stricture, main complication of this dangerous condition, is treated by pneumatic dilation and eventually by esophageal replacement. We aimed, through this study, to report the outcomes of esophageal pneumatic dilation complicating corrosive substance ingestion in children in a developing country.

Methods This cross-sectional study was performed on the population of pediatric patients with caustic esophageal stenosis between January 2005 and December 2020. All patients underwent pneumatic balloon dilation. A logistic regression model was built to predict the probability of the occurrence of the event (success/failure) of the dilation. The ROC curve is used to evaluate the performance of the logistic regression model to discriminate between positive and negative values of the dependent variable.

Results The success rate of pneumatic balloon dilation was 80.4%. The median duration of overall management was 11 months. The severity of caustic stricture observed during endoscopy was significantly linked to worse outcomes ($p = 0.001$). Multivariate analysis indicated that the severity of stenosis and the number of dilation sessions were independent risk factors for failure of dilation. ROC curve analysis showed that the area under the curve was 71.7%. A Cut-Off point value of 7 provided the best sensitivity and specificity.

Conclusion Pneumatic balloon dilation has been proven to be efficacious in infants with caustic esophageal stricture. Pediatric surgeons should take into account factors to promptly switch to replacement surgery and avoid unnecessary and time-consuming serial dilations.

Keywords Caustic · Esophageal stricture · Management · Balloon dilation · Outcomes

Corrosive substance ingestion (CSI) represents a significant public health issue due to its long-term health sequelae, as well as the associated economic and social burden on victims, families, and national healthcare resources [1–3]. Although industrialized countries have experienced a reduction in incidence rates, CSI is reported to be on the rise in developing nations due to a lack of regulatory framework, public awareness, and easy access to corrosive substances in households [2–6]. Children are particularly vulnerable to accidental caustic substance ingestion, accounting for 80% of all CSI victims [1]. In adolescents, however, such incidents are infrequent and often related to intentional self-harm [1, 2].

Acute complications of CSI are upper airway burn, mediastinitis, and perforation, which could be life threatening [7]. Dysphagia and esophageal strictures are found on the long-term follow-up.

Corrosive esophageal strictures (CES) formation typically occurs within three weeks of ingestion, and its

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severity depends on the nature and amount of corrosive agents ingested [7, 8]. While minor grade 1 [9] injuries have favorable prognosis, grade 2 and grade 3 injuries can lead to the development of strictures in 15% to 90% of cases, resulting in difficulties swallowing, abnormal esophageal transit, and malnutrition in affected infants, with an increased long-term risk of squamous cell esophageal carcinoma [7–11].

The treatment of CES involves medicines, endoscopic procedures, or surgery, based on the severity of the stricture and the overall health of the child [8, 11].

Currently, the mainstay of treatment for esophageal strictures is balloon catheter dilation. Surgical procedures such as esophagectomy or substitution with intestinal or gastric conduits are reserved for patients who do not respond to repeated dilations [11, 12].

In this framework, we aimed to assess the efficacy and safety of pneumatic balloon dilatation (PBD) for the management of corrosive esophageal strictures in the pediatric population and to identify potential prognostic factors that could predict the success or failure of PBD as a conservative treatment strategy and facilitate the timely decision to switch to esophageal replacement surgery.

Study design and methods

Study design

We performed a cross-sectional study on a population of pediatric patients diagnosed with caustic esophageal stenosis (CES), referred to the Pediatric Surgery Department during the period of January 2005 to December 2020.

Inclusion criteria

This study included all pediatric patients referred to the department of pediatric surgery, national referral of PBD, for the management of CES. The diagnosis of CES was confirmed by endoscopy or barium swallow findings. All included infants underwent either fluoroscopic or endoscopic PBD.

Exclusion criteria

Patients with active esophageal perforation were excluded from our study.

Data collection

This study involved a meticulous review of the medical records of patients in order to obtain pertinent sociodemographic and medical history data. The time of onset of clinical symptoms and identification of the caustic agent ingested by the patient were determined. Precise morphological features of the esophageal strictures were based on radiological and endoscopic findings. Relevant details of the pneumatic balloon dilation (PBD) procedure were also recorded, including the interval between ingestion and dilation, the number of sessions, the time intervals between sessions, complications related to PBD, and outcomes.

Pneumatic balloon dilation technique

The PBD procedure was performed following the diligent acquisition of informed consent from the parents or legal guardians of each patient. All endoscopic interventions were conducted under general anesthesia with endotracheal intubation to ensure the safety of the airway.

During the procedure, a pediatric rigid endoscope was introduced to the level of the stricture to determine the diameter of the narrowed esophageal lumen. The wire-guided balloon (Boston scientific Microvasive)), with varying diameters and lengths ranging from 8 to 20 mm and 5 to 8 cm, respectively, was selected based on the patient's age and the site of the stricture. Gradual inflation was achieved using a syringe pressure system, reaching pressures of 3 to 8 atm for 2–3 min before being deflated. After catheter removal, the esophagus was meticulously examined for any adverse effects, including bleeding, perforation, or widening of the narrowed area.

Patients were monitored for several hours for any signs of esophageal perforation and mediastinitis. Chest X-rays were performed to rule out any perforation before patients were allowed to resume oral feeding and discharged home.

Multiple dilations were performed every three weeks, with the frequency individualized based on the patient's clinical signs of dysphagia or endoscopic signs of stricture relief. For patients with recurrent strictures despite repeated dilations, local application of corticosteroids on the stenosis was employed as adjuvant therapy.

The favorable/successful outcomes of PBD were defined as patients regaining the ability to swallow solid foods. If dysphagia recurred despite repeated PBD, esophageal stenting (Boston scientific Endoprothesis) was performed to maintain an appropriate lumen and esophageal patency. In the case of very high or multiple stenosis that could not be stented, surgical reconstruction (esophageal replacement) was proposed.

Statistical analysis

Descriptive statistics were utilized to summarize the sociodemographic and clinical characteristics, as well as the radiological features of patients. The Kolmogorov–Smirnov test was used to evaluate the distribution of quantitative variables. The results were shown as a median and interquartile

range (IQR). Qualitative data were expressed as numbers or percentages.

Continuous variables were subjected to the Mann–Whitney *U* test. Categorical parameters, on the other hand, were evaluated using the Chi-square and Fisher's exact test. Multivariate logistic regression was employed to identify the factors that influenced the PBD outcomes, with the related odds ratios (OR) determined at a 95% confidence interval (95% CI). A logistic regression model was built to predict the probability of the occurrence of the event (success/failure) of the PBD based on explanatory variables. The ROC curve is then used to evaluate the performance of the logistic regression model in terms of its ability to discriminate between positive and negative values of the dependent variable. A p value < 0.05 (two-tailed) was deemed to be statistically significant in all analyses. IBM SPSS Statistics (version 21.0) was employed for all analyses.

Ethical considerations

This study involved the utilization of medical data obtained from the records of pediatric patients that were primarily collected for non-research purposes. To safeguard the ethical integrity of the investigation, the research protocol underwent rigorous review and received approval from the ethical committee of Fattouma Bourguiba University Hospital at Monastir-Tunisia.

Results

Baseline patient characteristics

A total of 107 patients with corrosive esophageal strictures (56 males and 51 females) were examined; Table 1 summarizes the demographic, clinical, imaging, and therapeutic features of the pediatric patients evaluated in our study. The caustic ingestion was mainly recorded in the age group of 2–5 years (median of 3 years). Alkaline substances were predominantly ingested ($n = 93$, 87%). Symptoms appeared on average 2 months (range 1–12 months) after ingestion of the corrosive substance.

Barium swallow analysis revealed at least one esophageal stenosis in all patients. The strictures were predominantly located in the upper and mid-esophagus in 41 (30.8%) and 23 (21.5%) of patients, respectively. Endoscopic findings categorized the strictures by severity into three groups defined by the diameter of esophageal lumen: moderately tight (> 5 mm) in eleven cases (10.4%), tight (3-5mm) in 44 cases (41%), or extremely restrictive (< 3 mm) in 52 cases (48.6%). Most of the strictures were long (2–5 cm) or very long (> 5 cm) in 55 (51.4%) and 33 (30.8%) of children, respectively.

Table 1 Demographic, clinical, imaging, and therapeutic features in our cohort

Sample size	$N = 107$
Age (months) Mdn [Q_{25}–Q_{75}]	36 [24–60]
Caustic substance *N* (%)	
Alkali	93(87)
Detergent	12 (11.2)
Acid	2 (1.8)
Dysphagia score *N* (%)	
Grade 1	9 (8.4)
Grade 2	33 (30.8)
Grade 3	42 (39.3)
Grade 4	23 (21.5)
Number of stenosis Mdn [Q_{25}–Q_{75}]	1 [1–3]
Stricture location *N* (%)	
Upper	41 (38.3)
Middle	23 (21.5)
Lower	11 (10.3)
Upper + middle	22 (20.6)
Middle + lower	10 (9.3)
Stricture severity *N* (%)	
Moderately tight	11 (10.4)
Tight	44 (41)
Extremely restrictive	52 (48.6)
Stricture length *N* (%)	
Short (< 2 cm)	19 (17.8)
Long (2–5 cm)	55 (51.4)
Very long (> 5 cm)	33 (48.6)
Delay caustic ingestion to 1st dilation (months) Mdn [Q_{25}–Q_{75}]	2 [2, 3]
Number of dilation sessions Mdn [Q_{25}–Q_{75}]	5 [3–9]
Management duration Mdn [Q_{25}–Q_{75}]	11 [6–20]

Mdn median, Q_{25} 25th percentile, Q_{75} 75th percentile

Endoscopic pneumatic balloon dilation (PBD) management

The management and outcomes of all patients treated with endoscopic PBD are detailed in Fig. 1. A total of 672 endoscopic dilation sessions were conducted over the observation period. The range between caustic substance ingestion and the first dilation exceeded 2 months in all cases. The median number of PBDs per patient was five sessions (range: 1–35). Less than six sessions were required for 53.3% of patients to recover normal feeding. Local corticosteroid application was performed in 60 cases (Fig. 1). The success rate of PBD was obtained in eighty six patients representing 80.4% of cases. The median duration of overall management was 11 months, with 67% of patients terminating sessions within one year.

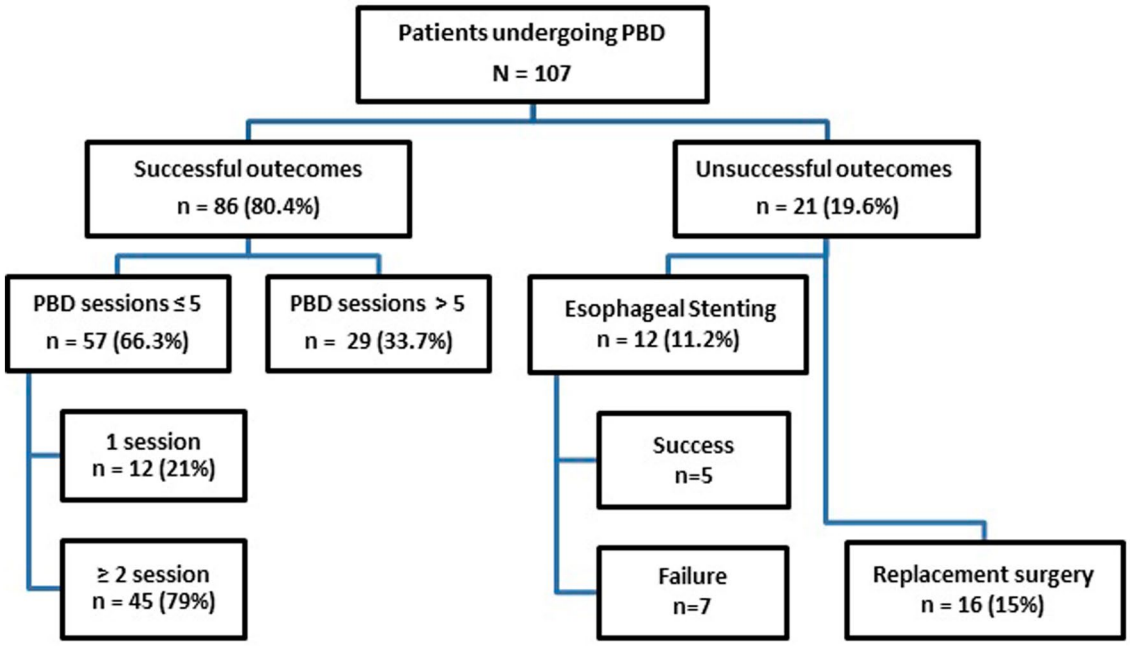

Fig. 1 Management and outcomes of all patients treated with endoscopic PBD

Our series reported a relatively high incidence of complications, including iatrogenic esophageal perforation ($n=41$, 38.3%) and gastroesophageal reflux ($n=10$, 9.3%) due to stenting. Five cases of esophageal perforation, confirmed via fluoroscopy during the dilatation session, resulted in secondary mediastinitis. In all cases, successful medical management (antibiotic associated to suction via a nasogastric tube and oxygenotherapy if needed) was achieved in esophageal perforations with favorable evolution and continued endoscopic dilations. Among children with gastroesophageal reflux, nine patients were treated medically, and only one required surgery (Nissen fundoplication). Endoscopic serial dilations failed to improve symptoms in twenty-one cases (19.6%). An esophageal replacement surgery was indicated in nine patients having a very long stricture. Twelve patients (11.2%) experienced recurrent strictures that required esophageal stenting. Stent migration occurred in six cases. A total of sixteen patients (15%) eventually underwent esophageal replacement surgery (colonic interposition), eight of whom had previously experienced esophageal perforation during PBD sessions.

Factors associated with endoscopic balloon dilation failure

We divided our sample into two groups based on the success or failure of endoscopic balloon dilation, as shown in Table 2. The severity of caustic stricture observed during endoscopy was significantly linked to worse outcomes ($p=0.001$). Long and very long strictures (> 2 cm) were also found to be significantly associated with endoscopic dilation failure ($p=0.018$). A greater number of dilations were significantly associated with successful outcomes ($p=0.011$). In our cohort, the occurrence of iatrogenic complications did not significantly influence the recovery rate.

Multivariate analysis indicated that the severity of stenosis ($p=0.002$, OR = 8.53, 95% CI 2.22–32.73) and the number of dilation sessions ($p=0.003$, OR = 1.14, 95% CI 1.04–1.25) were independent risk factors for pneumatic dilation failure.

The coefficient of determination between the discriminant function equation and endoscopic dilation outcomes was estimated at $R^2=0.285$.

The ROC curve was performed for this equation. The analysis of this curve revealed that the area under the curve (AUC) was good at 71.7%. A Cut-Off point value of 7 was selected, which provided the best sensitivity (%) and specificity (%). Results above this threshold indicated the failure of endoscopic serial dilation, while those below indicated its success (Fig. 2).

Discussion

Current management guidelines for esophageal strictures resulting from caustic ingestion in children recommend initial treatment with endoluminal dilation using either a push dilator (bougie) or a balloon dilator [13]. In recent years, the use of balloon dilators has become the preferred approach, performed endoscopically or over a guide wire with

Table 2 Factors associated with endoscopic balloon dilation failure

Variables	Endoscopic balloon dilation		*p* value
	Success (*N* = 86)	Failure (*N* = 21)	
Age (months) median [Q_{25}–Q_{75}]	36 [24–60]	36 [24–60]	0.676
Sex			
Male	47	9	0.332
Female	39	12	
Caustic substance			
Alkaline	76	17	0.281
No alkaline	10	4	
Dysphagia grade			
Grade 1–2	41	1	0.064
Grade 3–4	45	20	
Number of stenosis	1 [1, 2]	1 [1, 2]	0.672
Stricture location			
Upper	35	6	NA*
Middle	20	3	
Lower	7	4	
Upper + middle	19	3	
Middle + lower	5	5	
Stricture severity			
Moderate or tight	51	4	0.001
Extremely restrictive	35	17	
Stricture length			
Short ≤ 2cm	19	0	0.018
Long > 2cm	67	21	
Delay caustic ingestion to 1st dilation (months) Mdn [Q_{25}–Q_{75}]	2 [2–3.25]	2 [2, 3]	0.957
Local corticosteroid			
Yes	49	12	0.989
No	37	9	
Number of dilatation sessions Mdn [Q_{25}–Q_{75}]	5 [2.75–7]	9 [4–12]	0.011
Dilation-related complications			
Yes	40	11	0.629
No	46	10	
Iatrogenic perforation			
Yes	30	11	0.139
No	56	10	
Management duration Mdn [Q_{25}–Q_{75}]	11 [6–24]	12 [5–19]	0.997

Chi-square test: applicated

Mdn median, Q_{25} 25th percentile, Q_{75} 75th percentile

Non-applicated: Fisher's exact test

radiographic control [3]. This technique has been shown to be highly effective when performed correctly and promptly [12]. Balloon dilators come in various sizes, lengths, and designs, allowing for customization to the specific needs of each patient [12, 13].

In medical literature, endoscopic balloon dilation has reported success rates ranging from 64 to 100% in both adults and children, regardless of the underlying cause of the stricture [7, 8, 14, 15].

Our results further reinforce the effectiveness of using PBD for treating pediatric patients with CES. In a similar study conducted at a referral center in Egypt, El-Asmar et al. reported success rates of 98.2% for short corrosive strictures and 81.8% for long corrosive strictures following endoscopic balloon dilation (EBD) in 100 children [16].

While endoscopic dilation is an effective treatment for esophageal strictures, it is not without risks. Iatrogenic perforations are a known complication and have been

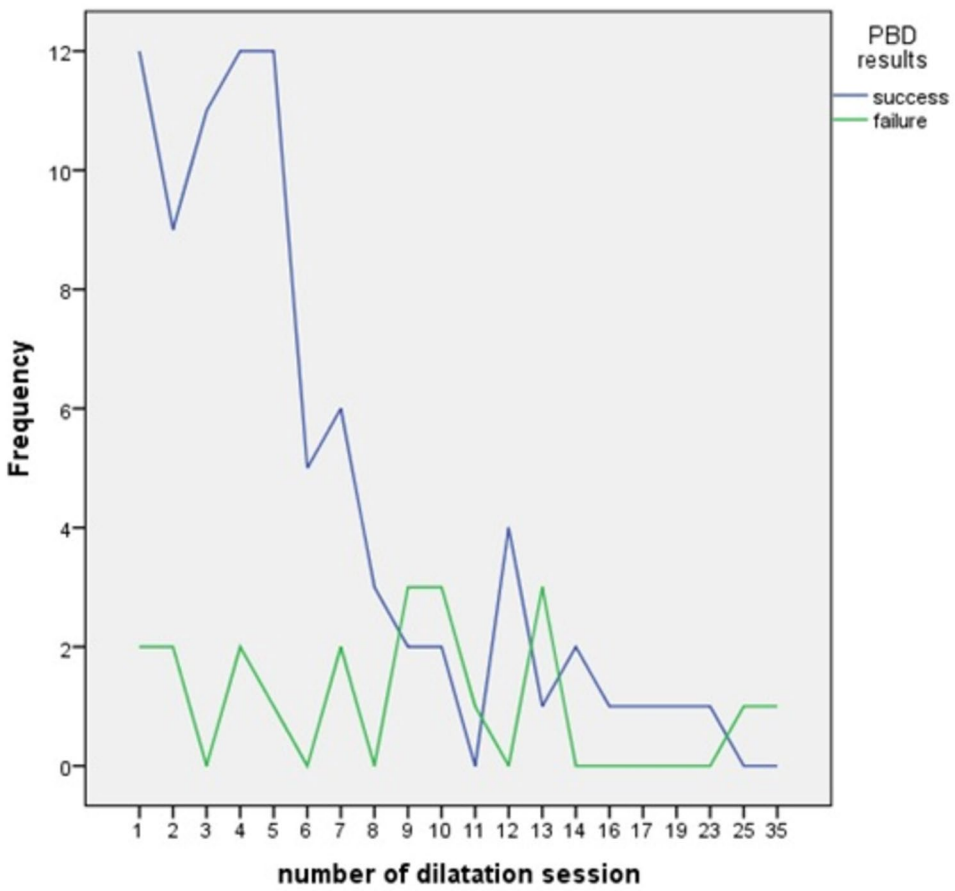

Fig. 2 Diagram of endoscopic PBD outcomes

reported in 0.6% to 18% of cases [7, 15, 17]. The incidence of iatrogenic perforation is higher in CES compared to congenital, peptic, or anastomotic strictures [5]. Research by Chang et al. demonstrated a 10% rate of iatrogenic perforation during endoscopic dilation, with 80% of cases occurring in infants with alkaline corrosive esophagitis [18]. This study suggested that alkaline corrosive esophagitis may increase the risk of perforation during EBD procedures due to severe esophageal fibrosis [8, 18, 19]. This may also explain the high rate of perforation in our patients, as many of them had ingested alkali agents.

In comparison to bougienage techniques, balloon dilators have been shown to be equally effective and safe [5]. Balloon dilation applies radial pressure to the stricture, which reduces the risk of esophageal perforation that may occur with bougienage techniques that apply shearing longitudinal forces [3, 12]. The choice between the two techniques should be based on factors such as the clinician's preference, local expertise, equipment availability, and cost-effectiveness [13]. Furthermore, fluoroscopically guided EBD provides superior image control, allowing for visualization of the entire esophagus, including its location, severity, length, rigidity, and shape, which can lead to increased treatment success compared to endoscopically guided EBD or blindly performed bougienage [20]. In our practice, we recommend fluoroscopically guided EBD for lengthy CES [21].

The achievement of desirable outcomes with EBD typically requires multiple sessions over an extended period [13]. Repeated sessions of endoluminal dilation may have significant negative social and psychological consequences on patients and their families, leading to prolonged absences from school and social activities. Deciding when to discontinue esophageal dilations remains primarily based on clinical judgment and experience with individual strictures, as no evidence-based criteria currently exist [13, 16, 22]. In this study, we aimed to identify predictive factors associated with the success or failure of endoluminal dilation to consider alternative treatments.

Similarly to previous studies, no statistically significant impact of age or gender discover any on EBD success [16]. Furthermore, our sample showed that the type of caustic agent ingested had no effect on the efficacy of EBD, which contrasts with the findings of Chang et al., who reported a significantly higher rate of EBD failure linked to alkaline corrosive injury [18].

Arlond et al. suggested that delaying stricture dilation initiation beyond three weeks from injury might worsen

fibrotic stenosis and prolong the need for dilation. They showed that children with stricture who received early dilation treatment (10–14 days) had highly satisfactory treatment outcomes compared to those who received late dilation treatment (≥ 21 days) [5]. However, we were unable to evaluate this factor in our study as our standard approach is to initiate dilations 4 weeks after CSI.

The literature has well documented that long, and grade 3 strictures have a lower response rate to dilations [3, 18]. El-Asmar et al. uncovered that the presence of long strictures (> 3 cm), pharyngeal extension, gastroesophageal reflux, and esophageal perforation during dilations were significantly associated with treatment failure. Furthermore, longer stricture lengths were found to have significantly longer cure times compared to shorter lengths [16]. Our study yielded similar results, as we identified grade 3 and long strictures as significantly associated with EDB failure. Our investigation also revealed a significant correlation between the total number of dilations and favorable treatment outcomes, contrary to the findings of previous studies [16].

Although some successful results have been obtained with dilation of esophageal stenosis with adjuvant local steroid (triamcinolone acetate) application, this approach is not validated and has not been prospectively examined in large series. The association of EBD with local steroid application in our practice did not significantly ameliorate the recovery rate.

In cases where patients have not shown significant improvement of dysphagia despite serial dilations or have factors that suggest failure of endoscopic balloon dilation, long-term stenting of esophageal strictures has been proposed as an alternative therapy [5, 23, 24]. The concept behind esophageal stenting is based on the same principle as dilations, whereby sustained circumferential radial pressure is applied to widen the lumen. Recently, self-expanding stents made of plastic, metal, and biodegradable materials have been utilized for pediatric patients with esophageal strictures or perforation [25]. However, the most common complication associated with stent placement is migration, which often necessitates stent removal or replacement [5, 26]. In a pediatric series, stent migration was reported in 48% of cases [23]. In our own experience, esophageal stenting was performed in 12 patients (11.2%), but symptom relief was unsuccessful in 58% of these cases.

Conservative management of CES may be considered unsuccessful if there is no sustained improvement in clinical symptoms and esophageal caliber within three to 6 months of therapy. Esophageal replacement is reserved for strictures that are excessively long or resistant to dilation. Some experts recommend a switch to replacement surgery after inadequate 12-week dilation therapy [7]. In pediatric patients, the commonly used techniques for replacement surgery include colonic interposition and gastric transposition operations [5, 27]. In our cohort, esophageal replacement was performed in 15% of children after failing serial dilation and/or stenting. This percentage appears to be higher than that reported by more conservative surgical teams, such as Karaman et al., who resorted to replacement therapy in only 5.7% of caustic strictures [8]. A colonic interposition with favorable outcomes was performed in all treated patients.

Following the conclusion of treatment for CES and the attainment of satisfactory relief from symptoms, it is imperative to conduct long-term surveillance on cured children in order to promptly detect any instances of secondary squamous cell esophageal adenocarcinoma. This form of carcinoma has been found to manifest between 1 and 3 decades after the corrosive ingestion and has a frequency at least 1000 times greater than that of patients in the same age group with no history of caustic substance ingestion [28]. It is essential to recognize that dilation, stenting, or esophageal replacement surgery undertaken after corrosive ingestion does not preclude the possibility of developing esophageal carcinoma [5, 7]. In our cohort, the mean follow-up of treated patients is relatively short, and we have not diagnosed any case of squamous cell carcinoma to date. However, we did offer families complete information on this issue.

This study presents a significant contribution to the understanding of the endoscopic and surgical management of CES in pediatric populations in our setting. With a large sample size, it provides a robust analysis of the efficacy and safety of the pneumatic balloon dilation technique. Moreover, it identifies several prognostic factors that can aid local healthcare providers in deciding when to discontinue conservative measures and switch to esophageal replacement surgery in pediatric patients.

However, the retrospective design of the study had limitations that made it difficult to identify potential confounding factors that could have influenced the results.

Conclusion

Our research provides valuable insights into the management of caustic esophageal strictures (CES) in pediatric populations using endoscopic pneumatic balloon dilation. This procedure has been proven to be efficacious and safe in infants. Pediatric surgeons should take into account factors such as the narrowness and length of strictures to promptly switch to replacement surgery and avoid unnecessary and time-consuming serial dilations. To optimize and standardize the endoscopic and surgical management of pediatric CES in Tunisia, further prospective and multicenter studies

are needed. These studies can help develop national guidelines, explore the use of adjuvant therapies, and minimize the risk of complications in (Tunisian) children.

Funding None.

Declarations

Disclosures Ben Youssef Sabrine, Marwa Messaoud, Belhassen Samia, Ben Fredj Meriem, Ben Salah Radhouane, Ben Mansour Maha, Chakroun Sawsen, Sfar Sami, Mosbahi sana, Ksia Amine, Gara Amel, Zemni Imen, Sahnoun Lassaad, Mekki Mongi, and Belghith Mohsen have no conflicts of interest or financial ties to disclose.

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