

R E V I E W

Endoscopic dilation in pediatric esophageal strictures: a literature review

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Summary. *Background:* Esophageal strictures in pediatric age are a quite common condition due to different etiologies. Esophageal strictures can be divided in congenital, acquired and functional. Clinical manifestations are similar and when symptoms arise, endoscopic dilation is the treatment of choice. Our aim was to consider the efficacy of this technique in pediatric population, through a wide review of the literature. *Method:* A search on PubMed/Medline was performed using “esophageal strictures”, “endoscopic dilations” and “children” as key words. Medline, Scopus, PubMed publisher and Google Scholar were searched as well. As inclusion criteria, we selected clinical studies describing dilations applied to all type of esophageal strictures in children. Papers referred to single etiology strictures dilations or to adult population only were excluded, as well as literature-review articles. *Results:* We found 17 studies from 1989 to 2018. Overall, 738 patients in pediatric age underwent dilation for esophageal strictures with fixed diameter push-type dilators (bougie dilators) and/or radial expanding balloon dilators. Severe complications were observed in 33/738 patients (4,5%) and perforation was the most frequent (29/33). Conversion to surgery occurred only in 16 patients (2,2%). *Conclusions:* Endoscopic dilation is the first-choice treatment of esophageal strictures, it can be considered a safe procedure in pediatric age. Both, fixed diameter push-type dilators and radial expanding balloon dilators, showed positive outcomes in term of clinical results and cases converted to surgery. However, it's essential to perform these procedure in specialized Centers by an experienced team, in order to reduce complications. (www.actabiomedica.it)

Key words: endoscopic dilation, pediatric endoscopy, Savary-Gilliard dilator, balloon dilator

Introduction

Esophageal strictures in pediatric age are a quite common condition, that may have different etiologies (1). In adults, esophageal tumors are the most common cause of strictures, while in children the etiological spectrum is broader (2). It is possible to distinguish among congenital forms, acquired forms and those deriving from functional disorders (achalasia) (1, 2). In congenital strictures, different subtypes have been described. The two most important are the fibro-

muscular subtype and the tracheal cartilaginous remnant subtype. In acquired forms, we can distinguish among caustic, anastomotic, peptic, actinic and neoplastic strictures. We can also identify strictures deriving from pathologies as epidermolysis bullosa and eosinophilic esophagitis (1). The most common causes are complications of surgical treatment of esophageal atresia, or esophageal burns due to caustic ingestion (3) that occurs especially in children of five years of age or younger (4), even though there are relevant variations from one country to another, especially be-

tween developed and developing countries in terms of incidence (2).

Failure to thrive is the most important consequence of this clinical condition, as it causes an impaired oral intake (5).

The endoscopic treatment of esophageal strictures has been reported to be the most frequent strategy in children (6). There is no universally accepted standard for the choice of the endoscopic technique in patients with esophageal strictures (3). Improvements in endoscopes and accessories have supported an increase in the number of patients who are conservatively treated with endoscopic dilations and a significant reduction of surgical treatments (6). Different dilators are now available. Fixed diameter push-type dilators as semirigid Savary-Giliard bougies and radial expanding balloon dilators that pass over a guide wire or through the channel of the endoscope are the most used devices, although there is still no consensus about which one has to be preferred (7). No prospective studies have directly compared the safety and efficacy of these types of dilators.

Esophageal dilation is associated with clearly defined morbidity and mortality and it should only be performed by experienced endoscopists, under general anesthesia (7). Perforation is the principal risk of this technique. The risk of this complication may be reduced by performing an accurate study of the stricture morphology and etiology, by choosing a correct type and size of the dilators and by performing dilations under fluoroscopic control (1).

The aim of this paper was to perform a review of the available literature on endoscopic dilations of esophageal strictures in pediatric age, with particular attention to possible complications and incidence to conversion to surgery.

Methods

In order to evaluate the efficacy and the safety of endoscopic dilations of esophageal strictures, we performed a literature search of PubMed database using the following key words "endoscopic dilations", "esophageal strictures", "children". Medline, Scopus, PubMed publisher and Google Scholar were searched as well. The entire databases were considered, without

restrictions of time. We included only full text papers selected with two filters "humans" and "language" (English papers).

We included all papers related to a pediatric population even if not exclusively.

Exclusion criteria were:

- papers referred only to single etiology strictures dilations
- papers referred to adult population exclusively
- study referred to other gastrointestinal tract strictures
- literature-review articles

Each article was tabulated in chronological order from the oldest to the most recent as follows: author and year of the study, number of patients, demographic data, endoscopic technique, total number of dilations, dilations for each patient, serious complications and conversion to surgery (Table 1). Regarding endoscopic techniques, we considered two different types: fixed diameter push-type dilators (bougie dilators) and radial expanding balloon dilators. Different adjuvant treatments were not considered.

The publications were manually screened and reviewed to identify reports and data were extracted from the papers according to the predetermined criteria. Two investigators independently reviewed and extracted data from the papers according to the predetermined criteria.

Results

We found at first 324 papers. Including only full text papers we limited the research at 234 study. Finally selecting two filters "humans" and "language" and including only English papers, we obtained 104 papers. After manual screening according to established criteria, 17 retrospective articles from 1989 to 2018 were selected.

Study population size among papers was very different, from a small cohort of 5 patients (8) up to the most numerous one with 125 patients (9). Overall, 738 patients in pediatric age (less than 18 years) underwent esophageal dilation. Only one study (10) considered a not exclusively pediatric population including people from 10 to 80 years (mean age 58 years).

Table 1. Articles included in the literature review

Author, year	N° patients	Demographic data	Endoscopic technique	N° of dilations	Dilation/patient	Serious Complications (total n° and %)	Conversion to surgery
Gandhi RP, 1989	12	<18 years	Tuckers	-	-	Yes 1 perforation (1%)	No
Shah, 1993	17	1 month- 15 years	Ballon dilation	132	7,7	Yes 1 perforation	Yes, 1
Jawad AJ, 1995	36	<18 years	Ballon dilation and Savary-Gilliard	-	-	Yes 1 perforation 1 anastomotic leak	Yes, 2
Wang YG, 2002	55 (40 M, 15 F)	10-80 years (median age 58)	Savary-Gilliard	401	7,2	No	No
Lan LC, 2003	77	2 months-20 years	Ballon dilation	260	3,3	Yes 4 perforation (1,5%)	Yes, 1
Bittencourt PF, 2006	125	1 month-16 years	Savary-Gilliard	-	-	Yes 5 perforation	No
Khanna S, 2008	5	4-12 years	Ballon dilation	-	-	No	No
Saleem MM, 2009	38	1 months- 10 years (median age 3,2)	Tuckers	801	21,1	Yes 2 perforation	Yes, 2
Alshammari J, 2011	49	<18 years	Ballon dilation	-	-	Yes 3 perforation	Yes, 6
Chang CF, 2011	10	1-50 months	Ballon dilation	-	-	No	Yes, 1
Lakhdar-Idrissi M, 2012	60	10 months-17 years	Savary-Gilliard	247	4,1	Yes 2 perforation	No
Shehata SM, 2012	38	5-22 months	Savary-Gilliard	654	17,2	Yes 1 perforation 2 small diverticulum	Yes, 3
Van der Zee D, 2014	19	1 month-15 years	Ballon dilation	87	4,5	No	No
Pieczarkowsky S 2016	106	1 month-18 years	Ballon dilation and Savary-Gilliard	347	3,2	Yes 1 perforation	No

(continued)

Table 1 (continued). Articles included in the literature review

Author, year	N° patients	Demographic data	Endoscopic technique	N° of dilations	Dilation/patient	Serious Complications (total n° and %)	Conversion to surgery
Cakmak M, 2016	38	0-14	Ballon dilation	-	-	Yes 4 perforation 1 fistula	No
Hsieh KH, 2017	10	<10 years	Ballon dilation	93	9,3	Yes 1 perforation (1%)	No
Al Sharkhy AA, 2018	43	2-17 years		180	4,1	Yes 3 perforation	No

Different endoscopic dilatation techniques were used: in 6 studies fixed diameter push-type dilators were preferred, in 2 Tuckers (2, 4) and in 4 Savary-Gillard were used (9-12); in 8 studies radial expanding balloon dilators were used (5, 8, 13-18); in 2 works (3, 4) a combination of two techniques was used, while in 1 study (19) the technique was not specified.

Even if in 7 papers (4, 5, 8, 9, 13, 14, 18, 20) the total number of dilations was not described, overall 3202 procedures were performed (median number 6,9/patient). Making a comparison between the two techniques, the median number of dilations with fixed diameter push-type dilators was 12,3/patient while it was 5,8/patient with radial expanding balloon dilators.

A total of 35 serious complications were recorded, 1 anastomotic leak, 2 small diverticulum formation, 1 fistula and 31 perforations. In 4 study complications were not described (8, 10, 14, 18).

Finally, in 10 studies (3, 5, 8-11, 15, 18-20) an eventual conversion to surgery was not described. In the other cases a total of 16 patients underwent surgical treatment, due to inefficacy of the dilation or for the management of the complication.

Discussion

Esophageal strictures in pediatric age are a quite common condition (1). In adults, the most common cause of strictures are esophageal tumors, while in

children the etiological spectrum is broader (2). In the present review only Wang et al in 2002 studied a wide aged population from 10 to 80 years (with a median age of 58). All 55 unselected consecutive patients were treated with Savary-Gillard bougies achieving in all cases a relief of symptoms. No serious procedure-induced complications occurred.

There is no universally accepted standard of endoscopic treatment of patients with esophageal strictures (3). In our review Gandhi et al. in 1989 and Saleem et al. in 2009 reported their experience with Tucker's string guide dilators while Wang et al in 2002, Bittencourt et al in 2006, Lakhdar-Idrissi et al and Shehata et al in 2012 described their one with Savary-Gilliard bougies. Eight authors from Shah in 1993 to Hsieh in 2017 used expanding balloon dilators while Jawad in 1995 and Pieczarkowsky in 2016 reported both, balloon dilators and semirigid Savary-Gilliard bougies. Overall, literature data show that there is no substantial preference between the two endoscopic techniques.

Comparing the efficacy of these two techniques, balloon dilators can be more effective and less traumatic than traditional bougies, as they provide a uniform radial force (16). More obviously, balloon dilation is performed under direct vision, furthermore the insertion of multiple devices is not necessary (8). In addition, endoscopic and fluoroscopic guidance allow a direct placement of the balloon catheter and visualization of the balloon inflation, thereby decreasing the risk of perforation (16). The only disadvantage is

that a balloon is single-use, therefore it is far more expensive than a bougie (8). If we consider number of dilatation per patient it is possible to observe how the median number of dilations with fixed diameter push-type dilators is higher than with radial expanding balloon dilators. Saleem in 2009 described a median of 21 dilations with Tucker's string guide dilators and Shehata in 2012 a median of 17 dilation with Savary-Gilliard bougies. Wang in 2002 reported a median of 7,2/patient with Savary-Gilliard, while Lakhdar-Idrissi in 2012 reported a better result with 4,1/patient. On the other side Lan in 2003 and Pieczarkowsky in 2016 reported similar results with a median number of 3,2/patient. Van der Zee in 2014 and Al Sharkhy in 2018 found similar values with respectively 4,5 and 4,1 median dilations per patient. Hsieh in 2017 showed slightly more numerous dilations, with 9,3/patient. Overall, the present review of literature confirms the superiority of pneumatic dilations compared to bougies.

Currently, esophageal dilation in children are almost exclusively performed under general anesthesia (3). Endoscopic esophageal dilation is associated with low risk of complications. No significant prognostic factors could be determined (13). The most frequent potential complication is bleeding and perforation is the most serious. Esophageal perforation remains the most dreaded complication for dilatation of esophageal strictures. A higher perforation rate has been esteemed for bougienage than for balloon dilation (16). The use of antibiotics is advised to reduce the potential complication of infection and more frequent scar formation in absence of antibiotic therapy (4). In literature, Wang in 2002, Swagata in 2008, Chang in 2011 and Van der Zee in 2014 did not report complications. Shah in 1993, Lan in 2003 et Hsiehin 2017 reported a similar rate of perforations with balloon dilators of 1-1,5% while Saleem in 2009, Lakhdar-Idrissi in 2012 and Shehata in 2012 reported a very low rate of perforations. Probably, the very low rate of complications in pediatric age can be explained with the constant practice of operative endoscopy in the operating room and under general anesthesia, to maximize safety.

The resort to surgery is a possible, even if uncommon, eventuality. Therefore, it should be reserved for those patients in whom endoscopic dilation has failed

and for those with complications caused by dilation (8) due to its association with high mortality rate and high complication rate.

Consecutive dilation procedures are recommended for at least 2 years before deciding their failure (2). Many authors recommend a six to 12-month period of conventional repeated esophageal dilatation (4) Determinant factors of success or failure vary in reported series and include: age, site of the stricture, tightness of stricture, length of stricture, number of strictures and failure to respond to dilatation (2).

In literature two studies, Alshammari and Chang both in 2011, needed surgery for dilation's failure. For the first author, 6 surgical interventions were described, 3 due to perforation and 3 for failure of the procedure, while for the second author a conversion to surgery was necessary in absence of complications.

Moreover, the review showed also a few surgical treatments of complications. Shah in 1993 described only a perforation treated by surgery while Jawad in 1995 and Saleem in 2009 reported two surgical treatment, for perforations and for anastomotic leak. Lan in 2003 had 4 perforation, but only in 1 case surgery was necessary, while Shehata in 2012 reported 3 surgical treatments, among whom, one perforation and 2 small diverticulum formations.

Overall, in literature a very low rate of need of surgery is reported and it is required mostly for the treatment of complications.

Conclusions

Esophageal dilatation represents a small percentage of pediatric endoscopic procedures (17) and it represents the first-choice treatment of esophageal strictures. Both, fixed diameter push-type dilators and radial expanding balloon dilators, showed positive outcomes in term of improvement of clinical conditions and cases converted to surgery, although the efficacy of pneumatic dilations seems to be superior compared to the use of bougies. Endoscopic esophageal dilation is associated with a low risk of complications. However, it is essential to perform these procedure in specialized Centers by experienced team, in order to reduce complications. Resort to surgery is a possible, even if

uncommon, eventuality and it should be reserved for failure and for complication's treatment.

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