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Research Article

Endoscopic Instrumental Dilatation of Benign Esophageal Stenosis in Children and Adolescents

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Received: December 25, 2017; **Published:** March 13, 2018

Abstract

Stenosis of the esophagus in children and adolescents can be either congenital or most often acquired. The symptomatology is related to the consequence of the stenosis and its complications may be dramatic, but by early diagnosis and treatment this can be avoided. The treatment is to ensure the continuity of the esophagus, by performing either an endoscopic instrumental dilation or a surgery of last resort.

Methods: this descriptive study including 63 young patients with esophageal stenosis were referred to us by the department of infant surgery for endoscopic dilatation. Their age ranged from 13 months to 16 years, and the sex ratio was 1.42 in favor of the male sex. The etiologies of the stenosis were listed as follows: caustic injury in 62% of cases, peptic in 22%, anastomotic stenosis in 11%, early postoperative recurrence of achalasia in two patients (3%), and one case (1,6%) of postoperative stenosis for treatment of gastroesophageal reflux (Toupet technique).

Prior to dilatation, a clinical-biological, pre-anesthetic, and esophago-gastric barium transit were performed to determine the patient's condition and the characteristics of the stenosis.

Endoscopic dilatation was realized with Savary-Gilliard bougies, a wire-guided balloon and/or a thrue the scope (TTS) balloon introduced into the operating channel of the endoscope, without systematic fluoroscopic control.

Results: A success rate was estimated around 68%; 12 patients (19%) had to be operated on failure of dilatation due to early and multiple recurrences of the stenosis, and finally we have had a case of instrumental perforation operated at the same time anesthetic. It should be noted that 7 (11%) patients never returned, we had classified them in the category of the lost of sight, but it is very likely that they answered the procedure well.

Conclusion: Endoscopic instrumental dilatation is an effective technique in the conservative treatment of esophageal stenosis, which must be used before surgery because of its high morbidity especially in the pediatric population. Other instrumental treatments have appeared in recent years, such as prostheses (metallic and plastic stents) that allow for extensive progressive dilatation and calibration of the stenotic zone of the esophagus. But the real treatment for acquired stenoses is prevention especially for caustic and peptic causes.

Keywords: *Endoscopy; Gastroesophageal Reflux; Esophageal Atresia; Caustic Stenosis; Peptic Stenosis*

Abbreviations

TTS: Thrue the Scope; EID: Endoscopic Instrumental Dilatation; PPI: Proton Pump Inhibitors; VTES: Very Tight Esophageal Strictures

Introduction

Esophageal stenosis is defined as abnormal narrowing of the esophageal lumen causing clinical signs such as dysphagia, chest pain, vomiting, undernutrition, and weight loss. Stenoses of the esophagus are from various origins [1,2]. When they are congenital, the symptoms appear early in childhood, they result from an aggression on the esophageal mucosa causing lesions evolving to an inflammatory thickening and eventually to fibrosis. Patients have high morbidity with severe consequences such as weight loss, undernutrition, food impingement that can causes fistula or perforation, or bronchial pneumopathy. Early diagnosis allows the child to have a correct development. Stenosis may be acquired, due to a mucosal aggression that can generate severe consequences.

The treatment aims to improve dysphagia, and prevent recurrence. Currently, it consists of either surgery or endoscopic instrumental treatment by dilatation, or the establishment of removable full-covered self-expandable intraluminal prosthesis; the last two are conservative treatments that have greatly reduced the need for surgery.

Esophagectomy with the placement of an intestinal interposition graft is a source of high morbidity, with uncertain long-term results, and should therefore be reserved only in the event of failure response to conservative treatment [3]. The endoscopist intervenes in order to ensure the continuity of the esophageal lumen by removing the obstacle in order to avoid complications and to prevent, if possible, recurrences.

The indications are to restore a normal flow of esophagus in children and adolescents in whom mutilating surgery could be catastrophic for their lives. The contraindications are the tight and long stenosis exceeding 8 cm, the presence of oesophageal fistula and finally a severe defect preventing the realization of the procedure under general anesthesia.

Endoscopic instrumental dilatation (EID) is an easy technique that should, however, be practiced only by trained endoscopists. Its morbidity is lower than surgery and the mortality almost nil.

Wire-guided dilatation is preferred for esophageal stenosis, it relieves the patient by greatly decreasing dysphagia.

Mechanical stenosis is fibrous and requires dilatation with bougies exerting a longitudinal force, while neurological stenosis such as achalasia due to lower esophageal sphincter hypertonia requires balloon dilation exerting radial force. There is therefore a choice concerning the instrument to be used for the dilation.

The most commonly used dilators are those guided on wire (Savary-Gilliard, Eder Puestow, Celestin, Plummer-Jackson), or on rubber wire (Tucker), and hydrostatic or pneumatic polyurethane balloons.

We wanted to report our experience in the endoscopic management of esophageal stenosis and to evaluate the causes and their response to endoscopic dilatation in children and adolescents who were entrusted to us by the department of infant surgery.

Materials and Methods

This is a descriptive study of 63 children and adolescents referred by the Department of Infant Surgery for endoscopic treatment of esophageal stenosis between 2008 and 2016. The patients were hospitalized in infant surgery, then prepared for the endoscopic treatment by performing a careful clinical examination, a radio-endoscopic assessment, and a pre-anesthetic assessment.

The clinical examination was to determine the symptoms, the general and nutritional status of the child and any complications. The degree of dysphagia was assessed by a clinical score [4]. Esophagoscopy and the contrast X-ray with barite ingestion was used to analyze the esophageal stenosis and assessed their location and diameter in order to select the appropriate dilation material. All patients were

subjected to a dilatation protocol performed under general anesthesia. The patients undergoing tracheal intubation and ventilation assisted and fasting for at least 6 hours.

Examinations were performed with Pediatric Olympus gastroscope (Olympus Corp, Tokyo, Japan). The material used for the dilation was either Savary-Gilliard bougies, a metal tracer guide wire (Wilson-Cook Medical Inc.), a wire-guided pneumatic balloon, or a TTS balloon inserted into the endoscope's operating channel.

Technique and protocol

The general anesthesia was performed by the resuscitation team using Nitrous oxide, Propofol and Midazolam. Then the paediatric endoscope is passed, the guide wire was placed under endoscopic without a systematic fluoroscopic guidance. For very tight esophageal strictures (VTES) in which the stricture is less than 6 mm, the tracer guide wire was used by us as a path-finder and also for Savary-Gilliard dilation with fluoroscopic guidance.

Different successive steps are followed during the instrumental dilatation technique: firstly, under endoscopic guidance, the stricture is approached. The metal guide wire is gently inserted through the stricture until the wire has been advanced more than 40 cm without strong resistance having been encountered; the fluoroscopy checking is only used with VTES.

Then after, keeping the wire in place, the scope is withdrawn; except when the stenosis is tight and short, particularly for the cervical site anastomotic stenosis, at this time we insert a balloon TTS and proceed to a pre-dilatation under endoscopic guidance.

The stricture is dilated over the guide wire starting with a 5 mm dilator using the markers on the wire and also on the dilators for guidance and under endoscopic control.

The dilatation sessions start 3 to 4 weeks following the ingestion in corrosive cases or other pathologies and in each single session, we did not use more than 3 sizes of dilators starting by the one suitable to the stricture caliber under dilatation and for each new size keep it at least for 3 successive sessions within three weeks period. Sessions were practiced every 2 to 3 weeks depending on the state of the esophagus. In one case, the dilatation was performed with a wire-guided balloon for the case of a post-operative recurrence of an achalasia.

After dilatation session, the patient remained under observation for at least 4 hours. A 6-hour nutritional diet was required, and feeding resumed in the absence of pain.

Dilatations continue till reaching the size of 11 or 13 mm dilator in first and second year of age, respectively, and 14 to 15 for the adolescents. When the caliber of 13 mm is reached, no more dilatation is needed, which is consistent with the upper normal limit for esophageal diameter in 2-year-old infant. In case of an insufficient response, when the progress to larger dilator sizes easily or resistance encountered, keep the size for further six successive sessions before going to the larger size.

We gradually space the frequency of dilatation by two to three weeks interval till it became once every three months for three successive sessions, which is considered the end of dilatation protocol to be followed after one year by endoscope or contrast radiography. Successful treatment was defined when the time interval between dilatation sessions is increased and increasing tolerance of age-appropriate food intake. Dysphagia score was done at 6 months after stoppage of dilatation for corrosive subgroup.

The ideal final diameter of the esophagus was determined by the symptoms reported by the patient and his nutritional status. The clinical parameters of good results of the procedure during the follow-up of the patients were the disappearance of the dysphagia and the weight gain, motivating the realization or not of other sessions of dilation. Patients no longer requiring endoscopic dilatation after six months were considered cured.

Clinical evaluation and contrast x-ray are performed 6 to 12 months after the last dilatation session. In case of recurrence of symptoms, further evaluation was done.

Heamorragia and perforation are the complications encountered during instrumental dilatation. The haemorrhage is often not serious, it is spontaneously resolving in the vast majority of cases. The mucosal dilacerations caused during dilation are systematic but they must not be deep because of the risk of perforation. If a perforation occurs, the treatment is endoscopic through the use of clips or surgical in the ultimate cases. The bacteremia caused by dilatation is a recognized complication. For this, an antibioprophylaxy is essential.

Results

We took care of 63 patients who were entrusted to us by the department of infant surgery for an endoscopic treatment of esophageal stenosis over a period of nine (09) years.

The age of the patients ranged from 13 months to 16 years. The calculated average was 5 years (Table 1).

The sex ratio was 1.42 (37 boys / 26 girls).

The age - sex distribution by cause shows the findings in the following table.

	Caustic	Peptic	Anastomotic	Other
Average Age	5,58 YO	4,75 YO	6,42 YO	9 YO
Sex : M	25	6	4	2
F	14	8	3	1

Table 1: *Distribution of the cause of stenoses by age and sex.*

The etiologies were represented as follows:

- Caustic ingestion was the most common cause: 39 cases representing nearly 62% of our patients.
- Peptic esophagitis in 14 cases (22%).
- Anastomotic stenosis in 7 cases (11%).

The etiologies were represented as follows:

- Two cases (3%) of early post-operative recurrence of achalasia.
- One case (1,6%) of post-operative stenosis following surgical treatment of gastroesophageal reflux (Toupet technique).

Characteristics of the stenosis: endoscopy was performed only after contrast radiographic of the esophagus, which provided essential information for subsequent management. These informations concerned:

- The site of the stenosis which was distributed as follows:
    - Cervical, represented 36% of stenoses (in 23 cases);
    - Thoracic, was 40% (25 cases);
    - Lower esophageal strictures accounted for 16% (10 cases);
    - And multiple stenoses (at least double) in 8% (5 cases).
- The character of the stenosis which:
    - Was simple and short in 44% (28 cases).
    - And complex (tortuous and/or long = 3 cm) in 40% (25 cases).
    - The diameter of the stenosis varied between 2 mm and 6 mm for an average of 3.4 mm.

Endoscopic instrumental treatment: all the patients were dilated under general anesthesia in the operating room in the presence of a team of intensive care physicians and anesthetists trained in the care of children.

The use of fluoroscopy is not regular. It is only used during the dilation of complex stenoses.

The material used in the various dilation sessions was:

- Savary-Gilliard bougies for caustic, peptic and sometimes anastomotic stenosis.
- Wire-guided balloons.
- TTS balloons that we used for certain cervical anastomotic stenoses.

Figure 1: *Dilation material used in our patients: a) Wire-guided balloon; b) balloon TTS with pump pressure gauge; c and d) Savary-Gilliard bougies.*

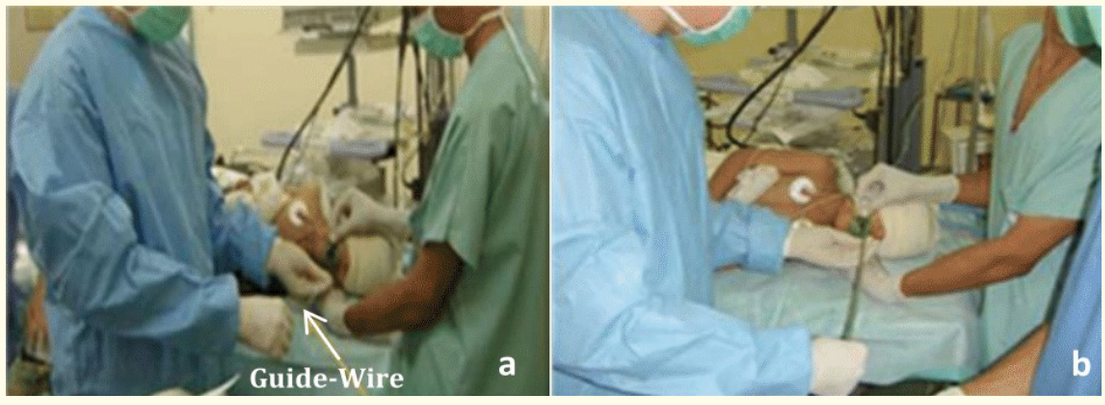

Figure 2: *Expansion technique: a) setting up the guide wire then; b) sliding the bougie on the guide wire.*

The number of dilation sessions varied from 1 session (recurrence of achalasia after surgery) to 10 sessions (Table 2 and figure 3).

Number of sessions [1 to 10]	Number of patients
1	5
2	11
3	8
4	6
5	12
6	3
7	5
8	3
9	4
10	3
1 then lost of follow	3
Average number of sessions per sick = 4.24	

Table 2: *Number of sessions per patients.*

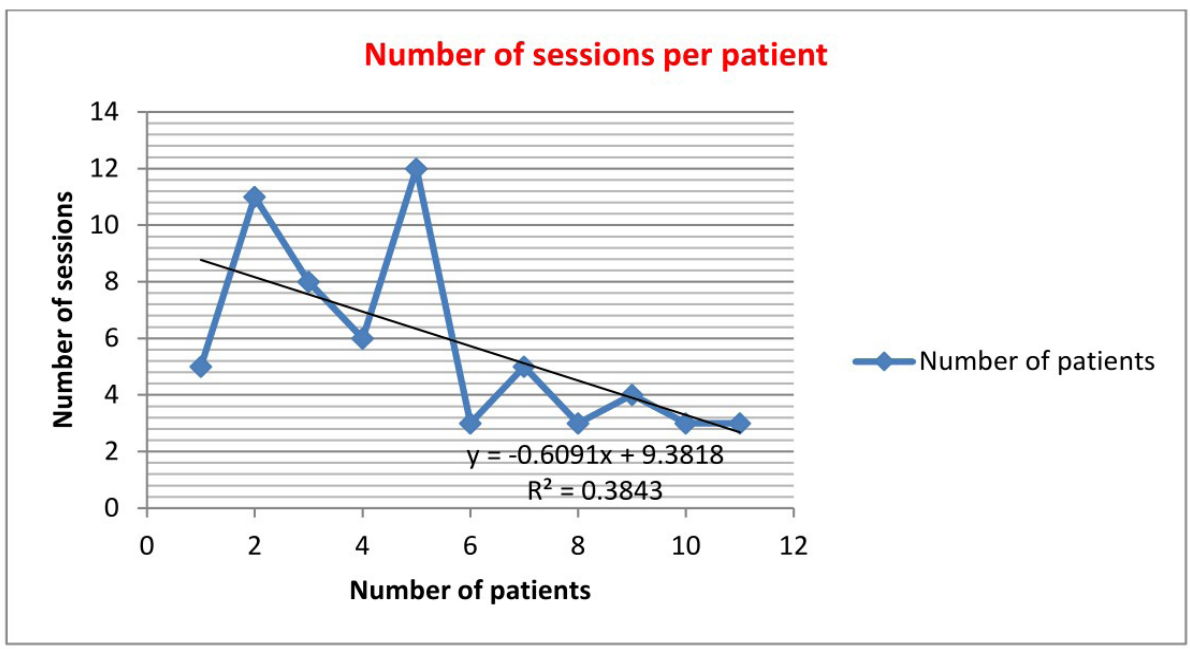

Figure 3: *Diagram describe number of sessions per patient.*

6 patients were not seen again after a single session. The average number of sessions per patient was 4.24.

The peptic etiology was the one in which the number of sessions was the most important. It took on average 6.25 sessions.

The caustic cause required on average 4.45 sessions and the anastomotic stenoses 2.85 sessions.

For dilatation, we used Savary-Gilliard bougies in 76% of cases, because of the fibrotic nature of the stenosis which was sometimes extensive. TTS and achalasia balloons were used twice. The combined use of bougies and the TTS balloon was performed in 10 patients (16% of cases).

Associated treatment: local corticosteroid treatment by intralesional injection, with the aim of reduces or even prevents recurrences, has only been carried out in a small number of patients and this did not allow us to draw any conclusion.

In addition, anti-secretory agents including proton pump inhibitors (PPI) and antibioprophylaxis were systematically instituted in all patients.

43 (68%) patients responded well to instrumental treatment by dilatation, while 12 (19%) had to be entrusted secondarily in surgery for failure of the dilatation. It should be noted that 7 (11%) patients never returned, we had classified them in the category of the lost of sight, but it is very likely that they answered the procedure well.

Morbidity: A case of perforation was found at the control immediate post-dilation, explained by the configuration of the bottom esophagus that had pronounced angulation in upstream of a tight peptic stenosis (Figure 4). It has been treated surgically at the same time anesthetic. We noted, moreover, 12 cases haemorrhage of small abundance spontaneously resolving, and no case of sepsis or other significant complication.

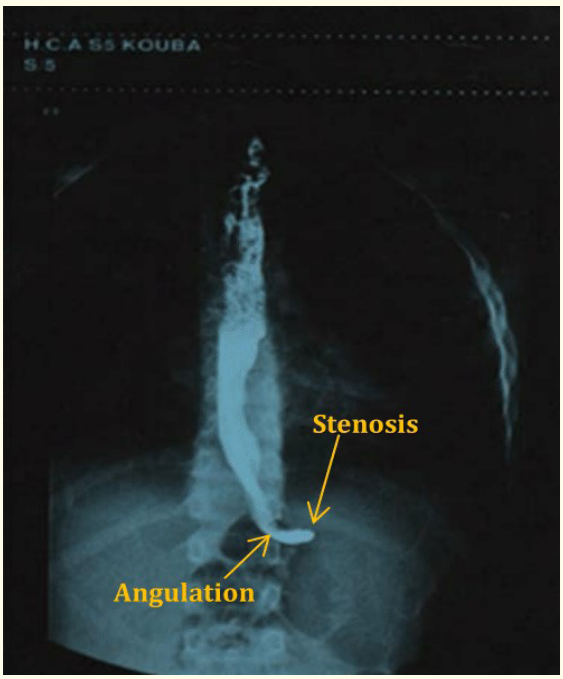

Figure 4: *Barium transit showing 90° angulation of the lower esophagus.*

Discussion

The incidence of esophageal stenosis in children and adolescents varies according to the geographical location and the therapeutic possibilities of each endoscopy unit [3]. The treatment of congenital malformations including esophageal atresia in newborns requires specialized care in intensive care unit by a medical and paramedical team well formed [5,6].

The increase in survival rate in these patients, as described in the literature, is correlated with the high number of therapeutic endoscopies treating anastomotic stenosis of the esophagus [10-12]. Acquired stenosis of the esophagus can be multiple. Secondary stenosis to the ingestion of caustic product remains, unfortunately, frequent in our country.

This is particularly the case in under developing countries where the application of preventive safety measures against the ingestion of caustics is not rigorous and mandatory [1,5-7].

In our case, it is particularly the lack of attention and insight of the parents who are at the origin of the accidents.

In our study, caustic esophageal stenoses represent 61% of all dilated stenoses, which is consistent with literature data for this subject. The stenosis can settle early, the EID must be practiced, according to the authors, enough early to 3 weeks or 1 month after ingestion. It must meet certain requirements such as: short stenoses, or long less than 5 cm. According to other authors, the endoscopic dilatation will be performed from 3 months once sclerosis is installed. Lesions of caustic esophagitis are readily available multiples affecting several segments, stenoses are often rigid, more extensive, technically more difficult to dilate, and have a higher recidivism rate compared to stenoses of other etiologies [3,5,8,9,14]. In our experience, we noted a number of larger dilation sessions for peptic stenosis followed closely by caustic etiology.

Gastroesophageal reflux disease is a common disorder in pediatric complicate of peptic esophagitis, however the prevalence of peptic stenosis remains poorly documented, it is estimated at about 1.5% [15,16]. Stenosis occurs after healing of a severe esophagitis, most often ulcerated. The preferred site of these stenoses is the third lower esophagus. Some predisposing factors have been reported such as disorders neurological, chronic pneumonitis, hiatal hernia, congenital malformation gastrointestinal or fundoplication failure [17,18]. These are the stenoses that respond the better at dilation. In our series, peptic esophagitis accounts for 22% of dilated stenoses.

In general practice, peptic stenosis responds well to instrumental dilatation when combined with appropriate anti-secretory treatment by PPI (complete resolution in 70 to 90% of cases). Poor healing and persistence lesions are a predictor of bad response to dilation [21]. A badly followed anti-secretory treatment is a source of recurrence of the peptic esophagitis even after total healing [19,20]. Nevertheless, it is known that fundoplication performed in children has a high rate of failure, for this the child must remain under medical treatment and endoscopic control after the procedure [20,21]. PPIs, because of their efficacy and tolerance, combined with endoscopic treatment should be the treatment of choice in children with peptic esophagitis complicated by stenosis, reserving surgical treatment only to refractory cases. For the diagnosis of GERD, there is mainly the clinic but in atypical cases pHmetry with or without impedancemetry can be indicated.

Anastomotic stenosis of the esophagus often has a better response to dilatation endoscopic instrument requiring less dilation sessions for an appropriate intraluminal diameter [5,6]. The etiopathogenesis of anastomotic stenosis is right now, badly understood. The genesis of the anastomosis is due to tissue hypoxia secondary to poor vascularization of the surrounding tissues, causing mucosal lesions followed by fibrosis.

We have, in our series, dilated 7 patients who had anastomotic stenosis. The average number of recurrences was 4.6 sessions per patient, which does not correspond to literature data. In refractory cases, with poor response to endoscopic dilatation, the risk of perforation of the esophagus during the procedure is higher. These cases often combine gastroesophageal reflux or congenital esophageal stenosis dysphagia [13].

Congenital stenosis of the esophagus (complete or incomplete atresia of the esophagus) is rare (1/25.000 to 50.000 births), and are secondary to the persistence of embryological vestiges tracheobronchial membrane, fibromuscular and cartilaginous tissues. Congenital cartilaginous stenoses must be operated because of the risk of perforation, whereas stenoses with a fibromuscular or membrane component are sensitive to endoscopic dilatation [22,23]. Endoscopic ultrasound is an indispensable diagnostic tool to guide the diagnostic. No cases are found in our series. Other causes of stenosis, less frequent, are represented by membranes and rings, ballistic injuries, road accidents or after voluntary ingestion or accidental foreign body. They can be iatrogenic secondary to a sclerotherapy of esophageal varices, elastic ligation, or after use of a laser or photodynamic therapy. Finally, certain real stenoses still remain of inaccurate etiology. With the recent appearance of the concept of eosinophilic esophagitis, certain criteria must, nevertheless, to be sought. We must think about it when there is a notion of food impaction, disorders esophageal motors, with or without mucosal lesions.

The diagnosis of esophageal stenosis is radio-endoscopic (Figure 4). The physician is first alerted by the patient's complaints, such as dysphagia, master symptom, and/or functional and physical signs related to the various complications.

The endoscopic dilatation and the stent placement are so-called conservative treatments, and their indication must be asked sparingly. Surgery should be reserved only for cases of failure of conservative treatment because of its high morbidity.

The instrumental dilatation of stenosis in our patients was achieved through the use of Savary-Gilliard bougies, pneumatic balloons whether they are wire-guided or introduced through the operator channel of the endoscope (TTS). Balloon dilation is effective by exerting a radial and uniform force on the stenosis, but was neither systematic nor the instrument of choice in our series. The cost and the unique use of balloons were limiting factors to their wide use. In addition, the literature shows no significant difference between wire-guided dilators and polyurethane balloons in terms of efficiency and rate of complications [5,10,12].

The dilatation must be secure in order to avoid complications. In our patients, the choice of dilatation equipment was done on a case-by-case basis, depending on the patient, the type of stenosis and its anatomical characteristics (site, diameter, length and number). The wire-guided balloon was used only in two patients in our study who had failed surgical treatment of an achalasia, and the TTS balloon combination and bougies was performed in 12 others, while the use of bougies alone has been used by all the rest of the patients. The prior use of a TTS balloon before that of the bougies was motivated by the cervical site of the stenosis and its very thin diameter, and the absence of fluoroscopic control; this allowed to dilate it slightly to allow access to the bougie. 3 successive sizes were used for the dilation and cannot exceed more than 3 times the initial diameter of the stenosis. However, in our work, especially in the short and tight fibrotic stenosis of the esophageal cervical site, a TTS balloon dilatation followed by bougienage was performed, and the result obtained was well satisfactory.

As a general rule, the procedure does not require the use of fluoroscopy, provided that the endoscopist strictly follows the rules of the procedure, its indications, and a good clinical and radiological evaluation of the patient. On the other hand, fluoroscopy, is essential in case of complex, tortuous, long or multiple stenosis, as well as to access difficulties of the guidewire [24].

The ideal esophageal diameter to be reached by dilatation is mostly determined by the patient, depending on the clinical context, improvement of dysphagia and/or nutritional status, weight gain but also the state of the esophageal mucosa in endoscopy [1,5,6].

The perforation of the esophagus is the most feared complication during instrumental endoscopic dilatation, its incidence is 0.7 to 3.5%, still considered a severe clinical situation [1,5,9,25]. In our series, only one case of perforation was noted (1.6%), recognized immediately after the endoscopic control procedure systematically performed. This complication had occurred because of the configuration of the lower esophagus which presented a 90° angulation, and the rise of the guide wire preventing the candle from sliding. Immediate restorative surgery was performed at the same anesthetic time, resulting in favorable suites. Some authors are in favor of a conservative treatment of the perforation of esophagus combining antibiotics, fasting, total parenteral nutrition, and clinical monitoring intensive [25].

Other endoscopic treatments, described in the literature, have been proposed during the stenosis of the esophagus. The intralesional use of corticosteroids (triamcinolone) is justified on the basis of reduced inflammatory response, and fibrosis, as well as restenosis after dilatation, motivated by relief of symptoms, maintaining a diameter of the esophagus during dilatation, and the increase in the interval between endoscopy sessions. However, the results are still heterogeneous and the procedure is not always sufficient to maintain the permeability of the esophageal lumen [1,26]. We have, in some of our patients, injected intralesional corticosteroids, motivated by the importance of the lesions, resulting in less recurrence but the few patients do not allow us to make any conclusion.

The development of self-expanding prostheses and removable plastic stents (Polyflex) allows introducing a new therapeutic weapon in the treatment of stenoses with the advantage of reducing the number of dilation sessions and maintaining the permeability of the esophageal lumen for longer periods, without the usual complications associated with metal stents [27]. Broto., *et al*. [7], in 2003, placed a stent in 10 children who had esophagus stenosis not responding to endoscopic dilatation, and performed well. This method seems to be effective, with low morbidity, we have unfortunately not had to use it for now but it should be in the near future.

Conclusion

This study shows that endoscopic dilatation of esophageal stenosis often gives good results and has a low complication rate.

Patients with caustic stenosis have higher morbidity and require more dilation sessions. These young patients must be treated case by case, even if the stenoses are of the same etiology.

It is extremely important that the staff caring for this type of patients should receive qualified training and be able to improve their skills in pediatric endoscopy, always bearing in mind the peculiarities of pediatric pathology.

However, despite current improvements in digestive endoscopy, prevention is still the best treatment for esophageal stenosis. Recommendations and precautions in the primary care of children should be able to provide an appropriate diagnosis, manage the taking into surgical load of congenital malformations, reduce the incidence of caustic ingestion, and finally optimize the medical treatment of gastro-esophageal reflux to avoid the development of complications.

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Volume 5 Issue 4 April 2018