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Endoscopic dilatation for benign oesophageal strictures in infants and toddlers: Experience of an expectant protocol from North African tertiary centre

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ABSTRACT

Purpose: The purpose of this study was to present the safety and effectiveness of an expectant protocol employing Savary-Gilliard dilatation in benign oesophageal stricture in infants and toddlers along a decade of experience. **Patients and Methods:** Thirty eight infants and toddlers with benign oesophageal stricture with age ranged between 5 and 22 months were treated by modified dilatation protocol. Complications and outcomes of the dilatation protocol were reported during a follow-up period of 12 to 100 months. **Results:** We have 25 cases of corrosive stricture, 4 congenital, 4 post-reflux and 5 post-anastomotic strictures. A total of 654 dilatations in 265 sessions were done. The corrosive subgroup has a mean dysphagia score of 2.6 pre-dilatation that improved at 6 months after end of dilatation to a mean of 0.3. In the non-corrosive subgroup, significant lower number of dilatation and sessions were reported. We have three failures that need surgery. Thirty five cases reach acceptable oesophageal caliber. Mucosal tear and small perforation each reported once. Small diverticulum reported twice. **Conclusions:** The expectant dilatation protocol is feasible in managing benign oesophageal stricture in infants and toddlers without increasing the morbidity. It is effective even in long segment or multiple corrosive strictures.

Key words: Benign oesophageal stricture, oesophageal dilatation, Savary-Gilliard dilators

INTRODUCTION

Benign oesophageal stricture is the common cause of stricture in infancy and childhood.[1] Dysphagia in infants and children differs from that in adults, and therefore requires special consideration.[1] Oesophageal strictures in children may develop as a primary congenital constriction, secondary to a surgically repaired oesophageal atresia with or without tracheoesophageal fistula, as a result of chemical injury after caustic ingestion, or following oesophageal surgery.[2-4] Gastro-oesophageal reflux and eosinophilic oeosophagitis represents another two aetiologies of rare incidence in infants. Dilatation of oesophageal strictures is a commonly performed procedure used to relieve dysphagia due to benign stenotic lesions.[3]

Little information is available regarding the safety and efficacy of dilatation of oesophageal strictures in infants and toddlers with Savary-Gilliard bougies.[5] This problem in infancy was reported in the literature as case reports or description of such group among older children' series.[1,4-7] Infants are more liable to complications than adults, despite that the majority is benign stricture.[5,6] Boyce's "role of three" has been postulated to decrease the morbidity following endoscopic dilatation.[3,5]

The optimal dilatation procedure is still controversial, especially in infants and young children.[8,9] Wang *et al.* document the safety of Savary-Gilliard dilators without the use of fluoroscopy for adult oesophageal stricture.[10] Most of the previous reports were concerned about the adult population mainly. Whether bougienage or balloon is preferable in benign stricture in children is an important issue need to be clarified in details.[11,12] The aim of this study was to describe our experience employing a modified expectant dilatation protocol using Savary-Gilliard dilators in the management of benign oesophageal strictures in infants and toddlers representing a special critical age group along a decade in a prospective manner.

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PATIENTS AND METHODS

This is a review of the outcome of dilatation of benign oesophageal strictures in 38 consecutive infants and toddlers with oesophageal strictures between January 2001 and December 2010 in our unit. Informed consent was obtained from the patients' parents and ethical clearance was obtained from ethical committee of Tanta Medical Faculty for the study.

The degree of dysphagia was assess using established dysphagia score.[5,11,13] Oesophagoscopy and barium swallow confirmed oesophageal strictures and evaluate their locations and diameters. All the patients were subjected to a modified dilatation protocol under general anaesthesia.

Examinations were performed with Pediatric Olympus gastroscope (Olympus Corp, Tokyo, Japan). Dilatation was performed with market polyvinyl Savary-Gilliard dilators and a modified soft metal tracer guide wire initially used for ERCP (300 cm length, with markers) (Wilson-Cook Medical Inc. Winston-Salem, NC, USA).

Technique and protocol

Under Ketamine anaesthesia, the paediatric endoscope is passed, the guide wire was placed under endoscopic and fluoroscopic guidance. For very tight oesophageal 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.

The technique is applied in the following steps: (1) under endoscopic guidance, the stricture is approached. The tracer soft guide wire is gently inserted through the stricture until the wire has been advanced more than 40 cm without strong resistance having been encountered and checked by fluoroscopy; (2) Keeping the wire in place, the scope is withdrawn; (3) The stricture is dilated over the tracer wire starting with a 5 mm dilator using the markers on the wire and also on the dilators for guidance and under endoscopic control; (4) We start dilatation 3 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; (5) If not responding well, in means that does not progress to larger dilator sizes easily or resistance encountered, keep the size for further six successive sessions to be nine in total (3×3), rather than only three representing the modification of the protocol to be more expectant in time before going to the larger size; (6) Dilatations continue till reaching the size of 11 or 13 mm dilator in first and second year of age, respectively. When the caliber of 13 mm is reached, no more dilatation is needed, which is consistent with the upper normal limit for oesophageal diameter in 2-year-old infant. This targeted caliber gave the chance after recoil and post-dilatation fibrosis to one size less oesophageal diameter enough luminal passage without dysphagia. In children, this is more accurate than the "rule of thumb" guide that is applied for adults. Then gradually, we space the frequency of dilatation by 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.

Figure 1 shows the steps of dilatation in a case of congenital stricture aged 9 months [Figure 1a-e], while Figure 2 showed the dilatation steps in a case of corrosive stricture aged 18 months [Figure 2a-f]. Failure was defined as abandonment of dilatation in favour of surgical intervention following non-progression for six months.

Follow-up of the cases was done and contrast radiography is performed 6 to 12 months after stoppage of dilatation. Further evaluation was done after the first year post-dilatation based on recurrence of symptoms at regular outpatient clinic visits. Results and number of dilatations of both corrosive and non-corrosive subgroups were compared and analysed using non-parametric Mann-Whitney test with significant probability value ≤ 0.05.

RESULTS

A total of 654 dilatations were done for the 38 infants and toddlers in 265 sessions. The sites of the strictures are summarised in Table 1. The success rate for both placement of the guide wire and dilatation was 100% with the use of fluoroscopy. In the non-corrosive subgroup, significant lower numbers of dilatations (10.23 *vs* 20.84 with *P* value of 0.001) and sessions (4.15 *vs* 8.44 with *P* value of 0.002) have been reported as compared to the corrosive subgroup.

Twenty three out of the twenty five corrosive cases have

long segment stricture where 5 of them were VTES where stricture is less than 6 mm.

Mild sepsis was reported twice with no further complications. Two cases developed small diverticulum in the congenital subgroup [Figure 1f] that gave no symptoms. Mucosal tear reported once that responded to conservative management, while small perforation occurred once that treated conservatively and dilatation resumed after 3 weeks with good response. No large full-thickness perforation or mortality was reported with this expectant protocol. There were no major complications among these infants.

Thirty three cases improved and reach a caliber equivalent to or near that of the normal oesophagus of age-matched infants, while 2 cases showed mild improvement with poor response. We have three failures that needed surgery, one post-corrosive case that was treated by gastric pull up and the other two were of congenital stricture cases that were treated by resection and primary anastomosis. One of these two cases of congenital stricture showed tracheobronchial remnant in the resected area, while the second case showed extensive fibrosis [Table 2].

There is improvement in the post-dilatation dysphagia score in corrosive subgroup as to be 0.3 *vs* 0.56 as compared to pre-procedure dysphagia score of 2.6 *vs* 0.64 with significant *P* value of <0.001. Table 2 depicted the response to dilatation as reported endoscopically at 6 months post-dilatation during the follow-up period.

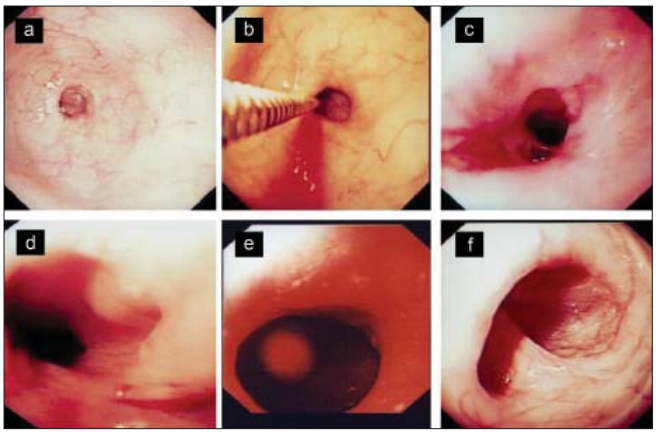


Figure 1: A panel of endoscopic photographs representing dilatation of congenital stricture in a 9-month-old infant where (a) Pre-dilatation photograph of congenital lower oesophageal stricture, (b) The same case with guide wire in place during a dilatation session, (c) The same case after dilatation and cutting the stricture at one side, (d) Endoscopic photograph of the same case 3 weeks after dilatation, (e) Photograph of the same case at cardia after passing the stricture site which is 1.5 cm above the cardia and (f) Endoscopic photograph of the same case shows post-dilatation diverticulum developed one year later

Post-dilatation contrast radiographic assessment was done and showed the excellent results of the modified protocol used as seen in Figures 3 and 4 in different types of strictures in infants and toddlers [Figures 3 and 4].

DISCUSSION

Oesophageal stricture in infants and/or toddlers is a challenging event facing paediatric surgeons or gastroenterologists. This problem is common in the third world countries where specialised centres are few. Successful oesophageal dilatation involves both successful placement of the guide wire and dilatation. This study demonstrates that Savary-Gilliard dilatation can be safe and successful with fluoroscopic control in infants, not only in children as reported earlier.[1,5] There were no serious procedure-induced complications. All cases were fed with liquid or semi-liquid diet formulae in the pre-dilatation period and two were fed through gastrostomy initially. Symptom improvement was achieved in all patients. Little information is available regarding the safety and efficacy of dilatation of oesophageal strictures in infants. Endoscopic and fluoroscopic guidance allow direct placement of the guide wire and dilators with visualisation of successive dilatation, thereby decreasing the risk of perforation in adults.[14] We reported similar results in oesophageal dilatation in infants and toddlers. Fleischer and others reported the need for fluoroscopic aid in almost 25% of his adult series and all cases of VTES,[10,14] but we use it exclusively in all cases as they are very susceptible and many of the corrosive cases were VTES.

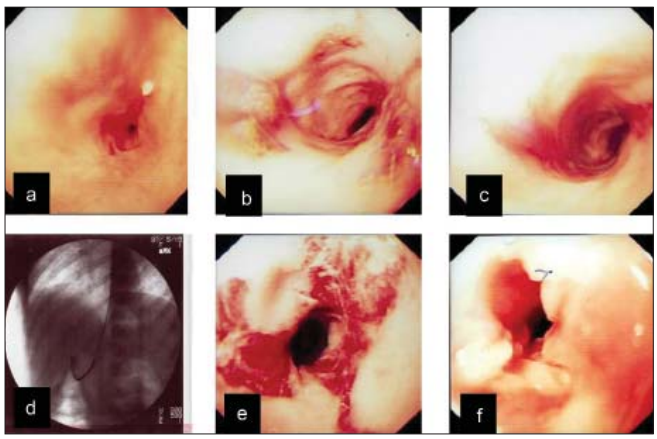


Figure 2: A panel of endoscopic photographs representing dilatation of corrosive stricture in a 18-month-old infant where (a) view of a VTES corrosive stricture, (b) Endoscopic view of the same case showed gradual dilatation at 3 weeks, (c) View of the same at 6 weeks, (d) Photograph showed the fluoroscopic guidance during the dilatation procedure with placement of guide wire, (e) Photograph of the same case at 3 months during dilatation procedure and (f) Endoscopic photograph of the same case 9 months post-dilatation

The best dilatation technique for a specific type of stricture is still controversial.[11,15] Whether the bougie dilators or balloon dilators are better remains a big issue in management of oesophageal strictures and is related to the experience of the treating doctor.[12,16] Many questions need to be answered in this respect. We try to give answers to some of these questions. Our modified protocol gives satisfactory results in infants and toddlers. In our study, we prefer using the bougie dilators rather than balloon dilators since the operator can feel the dilatation ongoing under his hand. The feeling of how much resistance is offered by the stricture, and therefore not proceeds forcefully. This point is of importance, since considerable percentage of our strictures were a result of caustic ingestion which are long and deep strictures. Another point is that we have long experience with bougie dilatation. This personal preference is in accordance to many former reports in adult or older children.[5,16,17] Contini *et al.* reported that dilatation with Savary bougies seems safer than with balloon catheters in corrosive cases.[18]

Table 1: The site of oesophageal stricture among the studied group in relation to the underlying pathology

Stricture	Upper	Middle	Lower	Multiple
Corrosive	3	10	6	6
Congenital	1		3	
Post-anastomotic	1	4		
Peptic			4	

Table 2: The response to the expectant dilatation protocol in our series in relation to the aetiology of stricture at 6 months post end of dilatation

Cause of stricture	Excellent	Fair	Poor	No
Corrosive	15	7	2	1
Congenital	2			2
Post-anastomotic	4	1		
Peptic	3	1		

Where Excellent: indicates we reached a dilator size equivalent to normal (size = 13 in 2 years old) in 3 successive sessions, Fair: indicates we did not go beyond a dilator size of one size below the expected normal size (size = 11 in 2 years old) in maximum of 9 successive sessions, Poor: indicates we did not go beyond a dilator size of two sizes below the expected normal size (size = 9 in 2 years old) in maximum of 9 successive sessions, and No response: indicates we did not go beyond a dilator size of three sizes below the expected normal size (size = 7 in 2 years old) in maximum of 9 successive sessions

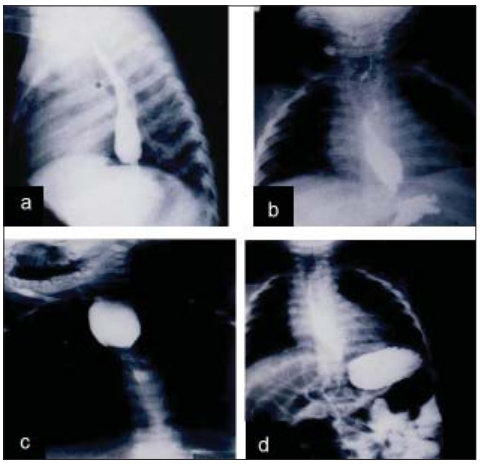


Figure 3: A panel of photographs of contrast X-rays where (a) Pre-dilatation oesophagogram in an infant showed lower congenital oesophageal stricture, (b) Post-dilatation oesophagogram of the same infant at 3 months after dilatation, (c) Pre-dilatation oesophagogram in an infant showed post-anastomotic stricture in an oesophageal atresia case in which the upper pouch is dilated and (d) Post-dilatation oesophagogram of the same infant at 6 months after dilatation

Despite many literature reports advocate the use of balloon dilatation to treat oesophageal stricture in children which is mainly non corrosive, yet it showed a considerable percentage of perforation.[19,20] Yeming *et al.* reported that the 3 failures among his series and the perforation occurred in children with caustic ingestion,[20] which is a further support to the results of our study since we had 25 out of 38 of our cases secondary to corrosive injuries. The expansion of this expectant protocol to 9 sessions rather than 3 sessions in cases that did not respond initially is the modification that gives satisfactory results in infants later. Although Poddar and Thapa stated that oesophageal perforation do occur in infants and children more frequently than previously reported,[5] our results using this expectant protocol document lower incidence of perforation in infants as compared to the older children in their series. In agreement to the results of our protocol, Hernandez *et al.* reported that polyvinyl bougie dilators have become

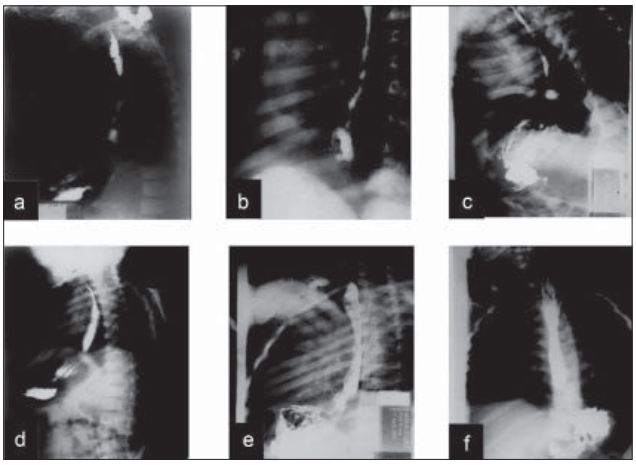


Figure 4: A panel of photographs of contrast X-rays of corrosive strictures in different group of infants where (a) Pre-dilatation oesophagogram of a corrosive case with long stricture at lower third of the oesophagus, (b) Pre-dilatation oesophagogram of another corrosive case showed multiple level strictures, (c) Pre-dilatation oesophagogram of a third corrosive case with mid third oesophageal stricture, (d) Post-dilatation oesophagogram of the infant A in the panel 6 months after dilatation, (e) Post-dilatation oesophagogram of the infant B in the panel 9 months after dilatation and (f) Post-dilatation oesophagogram of the infant C in the panel one year after dilatation

standard for more complex strictures in adults with the least perforation rate.[21,22] In our study and that of Poddar and Thapa,[5] bougie dilatation was employed. The common factor between both studies is that most of the cases are corrosive cases. This difference in the demographics of patients' group between these studies and different studies who advocate the use of balloon dilators as their main aetiology is non-corrosive, which are usually shorter and less deep strictures.[12,19,20]

The important point of our study group is the small patients' age. Expectant dilatation protocol hinders rushing in progression of dilatation, thus decreasing the complications and give better long-term results. We had only one case of mucosal tear and one case of small perforation that both treated conservatively. Perforation rate following dilatation in children was reported in previous studies as 2 out of 424 dilatations[17] and 2 out of 37 patients.[12] We reported perforation rate of 1 out of 654 dilatations, so the prolongation of dilatation time and number of sessions in our modified protocol is an advantage in decreasing the high incidence of morbidity and mortality in this delicate group of patients as reported in other studies with the conventional protocols.[12,16-19] In the current study, when we compared the dilatation in the same age group for non-corrosive strictures, we reported significant higher number of dilatations (20.84 *vs* 10.23 with *P* value of 0.001) and sessions (8.44 *vs* 4.15 with *P* value of 0.002) in the corrosive group. Also, Bittencourt *et al.* concluded that corrosive oesophageal strictures have a higher morbidity and require more dilatation sessions.[23]

Poddar and Thapa stated that dilatation should be terminated when resistance is encountered during three consecutive dilatations,[5] and this is not the case in our series with longer periods of dilatations, especially in corrosive cases. So, the results of our study suggest the non-validity of the former statement of Poddar and Thapa in long strictures (>3.0 cm), upper and subglottic caustic strictures. Obviously, in our series, long strictures or multiple strictures resolve under expectant dilatation protocol [Figure 4].

Many reports stated that endoscopic dilatation is considered to be the best treatment for most cases of benign oesophageal strictures.[1-3] Our results document this fact in infants and toddlers as a special age group even with long-segment and multiple corrosive strictures. Our results confirmed that the bougie dilatation is preferred in corrosive subgroup than balloon dilatation.[24] Symptom relief was achieved in all patients as seen by improvement of the dysphagia score in corrosive subgroup. Our results showed significant lowering of the dysphagia score after stoppage of dilatation by minimum of 6 months where the dysphagia score give lower value of 0.3 *vs* 0.56 post-procedure as compared to pre-procedure dysphagia score of 2.6 *vs* 0.64 with significant *P* value of <0.001.

Our good results in corrosive cases could be attributed also to the early bougien age in our protocol. This in accordance to Tiryaki *et al*. who reported that the strictures had resolved after 6 months of dilatation in patients initially treated with prophylactic early bougienage.[25]

In conclusion, the results of the current series showed that the expectant protocol we have proposed for oesophageal dilatation is feasible in the management of benign stricture in infants and toddlers, especially in caustic corrosive subgroup which is a very difficult one. We reported lower morbidity rate as compared to previous literature reports with conventional dilatation protocols.

This is one of the few studies concerning the infants' age group with benign oesophageal stricture to be managed by the Savary-Gilliard dilators.

This protocol gives satisfactory results with better quality of life when compared to surgery in those young infants with the known higher morbidity and mortality resulting from surgery.

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