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CLINICAL CASE

Caustic Esophageal Stenosis: A Case Report of Endoscopic Dilation With a Dynamic Stent

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KEYWORDS
Dilatation;
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Abstract
Introduction: The management of esophageal strictures has evolved from surgical treatment to the endoscopic dilation and, more recently, esophageal stenting.
Clinical case: We describe a case of a two-year-old boy with a double stenosis of the esophagus resulting from accidental ingestion of strong alkaline liquid. After several unsuccessful endoscopic dilations for three years and even topical mitomicin, it was decided to place a dynamic stent developed by the Digestive Surgery and Endoscopic Unit of the Bambino Gesù Hospital, Rome. The stent is a custom silicon device built coaxially on a nasogastric tube that is inserted after stricture dilations, by endoscopic guidance, and then fixed outside the nose. The device was removed after seven weeks with good clinical outcome (no dysphagia more than a year of follow-up).
Conclusion: This case confirms that the dynamic stent is a simple device that may avoid aggressive surgical substitution in cases of refractory strictures.

PALAVRAS-CHAVE
Dilatação;
Endoscopia Gastrointestinal;
Estenose Esofágica

Esofagite Cáustica Com Estenose: Um Caso Clínico de Dilatação Endoscópica Com Stent Dinâmico

Resumo
Introdução: O tratamento das estenoses esofágicas tem evoluído desde a correção cirúrgica, à dilatação endoscópica e, mais recentemente, à colocação de *stents* esofágicos.
Caso clínico: Descrevemos o caso de um doente que aos dois anos ingeriu acidentalmente um cáustico e desenvolveu duas estenoses esofágicas recidivantes. Após numerosas dilatações endoscópicas e aplicação tópica de mitomicina, ao fim de três anos, foi decidido colocar um

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stent dinâmico, desenvolvido pela Unidade de Cirurgia e Endoscopia do Hospital Bambino Gesù, Roma. O dispositivo consiste numa sonda nasogástrica com uma área de maior calibre (*stent)* que foi posicionada por via endoscópica na zona das estenoses e fixada por via nasal. O dispositivo foi retirado passadas sete semanas com melhoria clínica sustentada (ausência de disfagia mais de um ano após).
Conclusão: Este caso demonstra que o ''*stent*'' dinâmico é tecnicamente simples e permite evitar uma solução cirúrgica mutilante em casos de estenose esofágica de difícil controlo.

1. Introduction

Ingestion of corrosive substances remains an important public health problem mostly in developing countries. Children represent 80% of worldwide cases, primarily due to accidental ingestion.[1] The caustic substances may cause severe esophageal stenosis. The management of esophageal strictures has evolved and development of endoscopic techniques has led to a more conservative management, rather than more aggressive surgical substitution.

Endoscopic dilation (with balloon or bougienage) is used worldwide, although approximately one-third of patients develop recurrent strictures after dilation (defined as an inability to maintain a satisfactory luminal diameter for four weeks) and others have refractory strictures requiring multiple dilations (defined as inability to achieve a satisfactory luminal diameter over five sessions at two week interval).[2,3] On the other hand, endoscopic dilations have a significant risk of perforation (15–20%) and development of new strictures due to elevated pressure on the esophageal wall.[4–6]

In the last decade, esophageal stenting has become popular. Several authors have described their experience with different types of stents, although adapted pediatric devices are still scarce and the majority cause a centrifugal force on the esophageal wall while allowing the passage of food inside the stent. This type of stents may cause stretching and mucosal hypertrophy, with a non-negligible risk of migration, perforation and occlusion.[7–13]

Since 1988, the team of the Digestive Surgery and Endoscopy Unit of Bambino Gesù Hospital – Rome have developed a device named ''dynamic stent'' (DS), which allows food passage between the stent and the esophageal wall, improving esophageal motility and, thus, preventing stricture recurrence.[13,14] The DS is a custom silicon device built coaxially on a nasogastric tube to reach the desirable length and diameter, in order to create a larger area tailored to be placed along the stricture. The published experience of two decades of DS use confirms the safety and efficacy of the device and may even be considered as first option in the treatment of caustic esophageal stenosis.[14,15]

We describe a case of a patient with a refractory double caustic stenosis of the esophagus that was treated with a DS in the Unit of Pediatric Gastroenterology, Hospital Center of São João, Porto. Usually the caustic lesions are more severe at the site of functional narrowing of the esophagus (level of crossing the major vessels or just above cardia) and tend to be single with variable length, but on occasions there may occur at more than one site, as in the present case, causing additional therapeutic challenge.

2. Clinical case

A previously healthy two-year-old boy had accidental ingestion of strong alkaline liquid that led to a double stenosis (5 cm apart, at 15 and 20 cm from the mouth) of the esophagus. Each of the stenotic segments was short (less than 2 cm), but caused significant limited visibility and technical difficulty in addressing both for dilation. He was initially treated with endoscopic dilations with (Savary-Gilliard) bougies with recurrence of dysphagia two months later and even after subsequent other bougies dilation. On some occasions, when there was enough visibility of the esophageal lumen through both stenotic segments, TTS balloons of 10 mm were used, but once there was a perforation of the esophageal wall, successfully managed with conservative measures.

Several months after perforation, dysphagia recurred with grade 1 (dysphagia was graded using a previously described scale as follows: grade 0 = able to eat normal diet/no dysphagia; grade 1 = able to swallow some solid foods; grade 2 = able to swallow only semi-solid foods; grade 3 = able to swallow liquids only; grade 4 = total dysphagia),[16] that evolved to grade 3 twelve months later. Endoscopic dilations (initially with Savary-Gilliard bougies and after with TTS balloons) were resumed at two-week intervals. Topical mitomicin C was applied on four consecutive sessions initially at 0.1 mg/mL and then raised to 1 mg/mL. However, the stenosis remained refractory and multiple dilation sessions (more than 30, maximum width 12 mm) for 18 months.

Three years after the initial accident a DS was considered. Despite significant dysphagia (grade 3), the boy had adequate growth and weight gain mostly due to adjustment of food consistency to match his tolerance. Ethical clearance was obtained and parents provided informed consent. The exact location and extent of the stenosis were previously assessed radiologically with contrast (Fig. 1) and radio-opaque skin markers were applied. The two stenosis were then dilated until an adequate caliber was obtained (with Savary-Gilliard bougies of 7 and 9 mm followed by balloon of 12 mm) that allowed the placement of the stent (Fig. 2). The DS was then inserted through the mouth and the correct position was radiologically confirmed with the largest diameter at the level of the strictures using the skin markers as reference. Fig. 3 shows the actual device and scheme of the location of the device after insertion.