Case Report

Extensive caustic esophageal stricture in children can be treated by serial dilatations interspersed with silicone-covered nitinol stenting

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Abstract

Recurrent esophageal stenosis secondary to caustic ingestion may be challenging to treat. Self-expandable esophageal stents may be an alternative to repetitive endoscopic esophageal dilatation. We report a case of a 2-year-old male child with an extensive esophageal caustic stricture successfully treated using a combination of endoscopic dilatation and stenting. After 5 months of serial balloon dilatations, three nitinol internal silicone covered self-expandable stents were placed through the patient's gastrostomy spanning the entire esophagus. The stents were positioned using a combination of both endoscopic and fluoroscopic guidance. The procedure was repeated with only one stent 3 months later. A new stricture in the proximal esophagus needed surgical resection and anastomosis, followed by two pneumatic dilatations with progressively longer asymptomatic intervals. The results are promising with the patient able to use his own esophagus; however, this is a single case and optimal stent standing time is still to be determined.

Key words

Balloon dilatation, caustic ingestion, esophageal stenosis, self-expandable stents

Introduction

Management of corrosive ingestion and its sequelae constitute a challenging scenario. It is seen most often in children between one and three years of age, with boys accounting for 60% of cases. Self-expandable esophageal stents could be an alternative form of treatment to repetitive endoscopic esophageal dilatation.

Case Report

A 2-year-old male child was admitted to our center presenting an esophageal stenosis for 3 months, following an accidental

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caustic ingestion. The barium esophagogram showed a long stenosis from the cricopharyngeal level to the lower third of the esophagus [Figure 1a]. A retrograde esophageal dilatation program through a gastrostomy was repeated every 2 weeks (10 sessions in total) without improvement in swallowing. Five months after the injury, three nitinol internal silicone fully covered self-expandable stents (two of $16 \text{ mm} \times 60 \text{ mm}$ and one of $16 \text{ mm} \times 40 \text{ mm}$) were placed, resulting in full esophageal dilatation. These were placed starting with the most cephalad, ensuring 1-2 cm overlap between the stents to avoid displacement [Figures 2 and 3].

The patient presented with severe pain initially, following stent placement which spontaneously resolved after 4 days. He tolerated full feeding by mouth and there were none of

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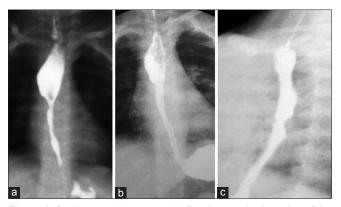


Figure 1: Barium esophagogram. (a) Previous to the insertion of the nitinol internal silicone covered self-expandable stents. Maximum esophageal filling. (b and c) 5 months later. Normal oral feeding (without dysphagia) after removal of the stents

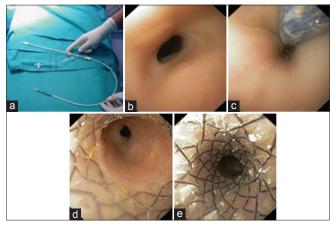


Figure 2: (a) Nitinol (nickel/titanium) mesh with smooth silicone inner surface and delivery. (b) Esophageal stricture (endoscopic view). (c-e) Stents positioning endoscopically and radiologically controlled

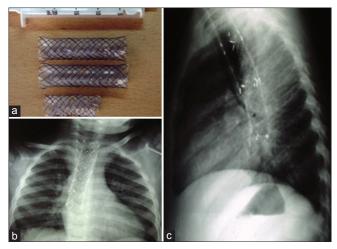


Figure 3: (a) Nitinol (nickel/titanium) mesh and smooth silicone inner surface. (b) Radiological image of the three inserted stents (anteroposterior view). (c) Radiological image of the three inserted stents (lateral view)

the possible early complications such as stent migration. Omeprazole (15 mg/24 h PO) was added to the treatment.

The stents were removed after 4 weeks, and one dose of mitomycin (0.4 ml/kg) was sprayed on the dilated area during the same procedure. Two residual 1 cm long stenoses were identified at the level of the cricopharyngeal sphincter and 3 cm above the gastroesophageal junction, respectively [Figure 1b and c]. The former was due to difficulty in stent placement with tracheal compression at the thoracic outlet area, and it was readily seen at the first stenting attempt. Three months later, another stent (16 mm \times 40 mm) was required, obtaining full dilation of the residual distal esophageal stenosis.

Balloon dilatations were done at intervals of 4 weeks, obtaining an elastic esophagus with normal appearance of the mucosa. The stricture of the proximal esophagus, however, had a poor response requiring surgical resection of the cicatricial tissue and anastomosis by left lower neck approach. This was followed by pneumatic dilatations which were initially bisemanal with progressively longer intervals over a 5 months period (7 sessions were performed). After 2 years of follow-up, the patient was fully asymptomatic without major abnormalities on esophagoscopy and barium esophagogram images.

Discussion

Ingestion of basic substances causes greater injury to the esophagus, specially affecting the areas of natural constrictions, when compared to acid.^[1]

The mainstay of treatment for esophageal stricture is dilatation with a 58%–96% success rate. [2] Results are better for patients with esophageal atresia in comparison with those with peptic or caustic strictures. Between 1 and 15 dilatations are required to treat symptomatic strictures, with perforation being the most serious life-threatening complication at an incidence of 0.1 and 0.4%. [3]

Mitomycin is an antibiotic and antiproliferative agent with antineoplastic properties produced by *Streptomyces caespitosus*. Its use shows significant promise for symptom-free intervals after dilations.^[4] A single dose of mitomycin was used with the patient instead of the literature recommended five doses, and therefore, its success as a form of treatment cannot be commented on in this case.^[5]

Temporary stenting with nonmetallic expandable stents can be effective for management of refractory, benign esophageal strictures when medical and endoscopic treatments fail. Many commercially available esophageal stents due to their size are unsuitable for use in pediatric patients. Custom dynamic stents^[6] and airway stents (off-label) have been used in children.^[7] The duration of stenting has not been determined, with a variable range of 4–6 weeks. Despite a limited number of pediatric studies, the clinical success rate with stents is 50%–85% for refractory, benign esophageal strictures.^[4] Self-expanding plastic stents (SEPS) seem to have good long-term results in

children but associated with an increased tendency to migrate. This complication is less frequent in self-expanding metal stents (SEMS) such as nitinol stents, for which the extraction procedure may be more traumatic.^[8,9]

According to published studies for adults, fully covered SEMS (coated with silicone) may be associated with fewer complications usually seen with partially or completely uncovered SEMS such as bleeding, fistulae, recurrent or new strictures, embedment, and erosion. Finally, although biodegradable stents are still associated with the complication of migration, stricture recurrence, and tissue ingrowth, preliminary data in adults suggest that biodegradable stents might provide a valuable alternative to SEPS and SEMS.^[9]

We placed nitinol internal silicone fully covered self-expandable stents in our patient, which were originally designed for tracheal stenosis dilatation. The reasons were its advanced technical design and big experience in adults. Dilatations needed after the stent placement were related to longer asymptomatic periods, suggesting fewer repeats of esophageal dilatations. Even with a refractory stricture, it is thought that stenting allowed for better tissue quality for the surgical resection and anastomosis.

Conclusion

While stenting is an established method of treatment of esophageal strictures in adults, only a few case reports describe the use of this therapy for children refractory to standard dilation. We successfully treated a 2-year-old male child using nitinol internal silicone fully covered self-expandable

stents with interval balloon dilatations. Endoscopic and radiologic control is essential for a precise stent positioning. The results obtained are promising although optimal stent standing time must be determined.

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Conflicts of interest

There are no conflicts of interest.

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