Bio-degradable stents – a new approach to the treatment of caustic stenoses in children

O. V. Bychkova, I. I. Lazyuk, V. Averin

Children's Surgical Centre, 1st Municipal Clinical Hospital, Medical University, Minsk, Belarus

Summary

Aim of the study: *To report on a 4-year-old boy with a corrosive oesophageal stenosis, in whom two subsequent SX-ELLA Oesophageal Biodegradable BD-Stents were successfully implanted.* Methods: *We describe the course of the caustic oesophageal stenosis experienced by the individual patient within a 2-year implantation period.* Results: *A 4-year-old boy accidentally swallowed caustic liquid and developed severe stricture lesions of the oesophagus which were repeatedly treated by pneumatic dilation for one year. Therapy was associated with serious complication such as oesophageal perforation, mediastinitis and pleuritis. With respect to the resistance of the stenosis to the standard therapy, esophagoplasty was considered as the only therapeutic option. Finally, a new SX-ELLA Esophageal Biodegradable BD-Stent (ELLA-CS s.r.o., Czech Republic) was implanted in November 2006. The implantation produced immediate relief from dysphagia, i.e. the score of 3 dropped to 0. The stent preserved its integrity for a period of 11 months after the implantation and evoked extensive mucosal hyperplasia and dysphagia. Dysphagia was successfully managed by repeated dilations within a 4-month period. The BD Stent fully disintegrated 18 weeks after implantation. The patient started to take esomeprazole. The interval between dilations extended to four weeks. However, the stricture got worse and had to be treated by implantation of a second BD Stent in December 2007, i.e. 13 month after the first implantation. The patient experienced a trouble-free period lasting 12 weeks. Then re-stenosis causing dysphagia developed above the proximal stent end. The complication was successfully managed by dilations within 4-week intervals. Disappearance of the original axial hiatal hernia and gastro-oesophageal reflux resulted in cessation of PPI therapy in June 2008. This was followed by development of a severe stricture formed in the middle oesophagus one month later. Esomeprazole administration was renewed and the interval between dilations increased to three weeks. In May 2009, i.e. 2.5 year and 1.5 year after the first and second BD Stent implantation respectively, the patient is exhibiting moderate physical development. He experienced a growth gain of 10 cm and weight gain of 20 kg. Although the BD Stent implantation was associated with some complications related to mucosal hyperplasia, we consider the therapy successful and are planning implantation of the next BD Stent. The advantage of the BD Stent is obvious compared to the potentially devastating effect of oesophagoplasty which had been originally considered the only therapeutic option.* Conclusions: *Although clinical experience is very limited, it seems that the BD Stent might represent a valuable therapeutic option in child patients with caustic stenosis intractable using standard dilation therapy. Basing on the medical history of the treated child, we learnt some principles to be followed in terms of therapy using the BD Stent.*

Key words: *caustic ingestion, corrosive oesophagitis and corrosive stenosis in children, bio-degradable stent*

Souhrn

Biodegradabilní stenty – nový přístup v léčbě kaustických stenóz u dětí

Tato kazuistika popisuje případ čtyřletého chlapce s korozivní stenózou jícnu, u kterého byly úspěšně postupně zavedeny dva biodegradabilní stenty (SX-ELLA Oesophageal BD-Stent). Čtyřletý chlapec náhodně vypil žíravinu, následně se vyvinula závažná striktura jícnu, která byla po dobu jednoho roku léčena opakovanými balónkovými dilatacemi. Léčba byla spojena se závažnými komplikacemi (perforace jícnu, mediastinitis, pleuritis). Vzhledem k refrakternosti stenózy na standardní léčbu, byla zvažována chirurgická plastika jícnu. Navrhovanou operaci rodiče chlapce odmítli. Proto byl v listopadu 2006 zaveden biodegradabilní stent (SX-ELLA Esophageal Biodegradable BD-Stent, ELLA-CS s.r.o., Hradec Králové). Stent přinesl okamžitou úlevu dysfagie. Stent zůstal in situ bez dezintegrace 11 měsíců, vyvolal výraznou slizniční hyperplazii, která vedla k novému objevení se dysfagie. Proto byl v prosinci 2007 zaveden nový biodegradabilní stent. V květnu 2009 (tj. 1,5 roku po zavedení druhého biodegradabilního stentu) je dítě bez větších potíží, za sledované období přibralo 20 kg a vyrostlo o 10 cm. Biodegradabilní stenty představují novou léčebnou možnost v léčbě refrakterních kaustických stenóz u dětí.

Klíčová slova: *biodegradabilní stent, korozivní ezofagitida, kaustická stenóza jícnu*

Accidental ingestion of a concentrated corrosive produces a high degree of caustic oesophageal lesions, resulting in severe extensive oesophageal stricture. Balloon dilation which is considered to be the first line of therapy does not always bring about the desired, long-term results. With respect to the poor recovery prognosis, these patients become candidates for oesophagoplasty. Oesophagoplasty with a segment of large or small intestine in children is associated with a high rate of complications. The impact of the method has to be considered with

respect to the patient's age. As for young children, we should take into consideration the impact of surgical treatment on bowel function and particularly on the organ growth.

From November 2006 to November 2008, Dr Jan Danis from Austria implanted four polydioxanone biodegradable oesophageal stents (ELLA-CS, Czech Republic) as an alternative to standard treatment methods in three children with caustic oesophageal stricture at the Children's Surgical Centre (Minsk, Belarus). We report the course of the disease in one of the children both before and after implantation of the stent. We subsequently implanted two biodegradable (BD) stents, knitted from polydioxanone fibre, into a 5-year-old patient who had experienced severe caustic oesophageal stricture resistant to standard dilation therapy lasting for one year. From November 2006 to August 2008, we followed up the effect of stent implantation, i.e. stricture dilation (change of stricture morphology demonstrated by oesophageal fluoroscopy), its clinical response (dysphagia relief) and gradual stent disintegration.

CASE REPORT

A 4-year-old boy accidentally swallowed caustic liquid containing a high concentration of sodium hydroxide in November 2005. Two weeks after the incident, the patient was admitted to the Children's Surgical Centre (Minsk, Belarus) in a serious condition. He was able to swallow only water (dysphagia score 3) and weighed 9 kg. Endoscopic examination revealed severe caustic lesion along the entire length of the oesophagus as well as a gastric lesion. Dilations were performed every 2–3 days within the first month. Later, the interval between dilation increased to 14 days. After 10 months of dilation therapy, the patient experienced an oesophageal perforation. He developed severe mediastinitis and pleuritis managed by thoracoscopic pleurodesis and drainage of the pleural cavity. After managing the condition, conservative treatment continued. The patient exhibited signs of toxic liver damage due to frequently performed anaesthesia (64 during one year) and first-grade iron-deficiency anaemia. The child was considered to be eligible for oesophagocoloplasty. The parents declined surgery.

In November 2006, when the attending physicians learnt of an alternative to the existing treatment methods, they decided to implant a biodegradable stent manufactured by ELLA-CS. At the time of stent placement, the patient suffered two oesophageal strictures (Fig. 1). The less obvious one was in the upper third of the oesophagus at the Th3 level; the second one was obvious and located at the Th6–8 level. Scar development led to oesophageal shortening, axial hernia and gastric angle deformation. In order to prevent potential wedging of the upper stent inside the lower stent we decided to place one stent into the tight stricture in the lower part of oesophagus. The patient underwent the stenting procedure performed under both endoscopic and fluoroscopic control in a satisfactory manner. During the two days after the operation, the patient was medicated with analgetics, antiemetics and broad-spectrum antibiotics. Later, the patient was not administered any medication. Fluoroscopic dynamic examination of the oesophageal motility as well as fluoroscopic control of the stent position was performed. Barium swallow control was performed 24 hours after implantation and within a 7-day period during the first month after implantation. The stent remained in-situ and dilation of the oesophageal stricture allowed free passage of food. Dysphagia was not present (dysphagia score 0) and the patient ate solid food. Control endoscopic examination was performed after two months (January 2007) and revealed extensive mucosal hyperplasia at the location of the stent, especially at its proximal part (Fig. 2). Resection of the hyperplastic oesophageal mucosa above the proximal stent end was performed. After 11 weeks (February 2007), when the stent was still in place (both radiopaque markers and stent mesh were kept), extensive oesophageal stenosis above the proximal stent end was observed. This situation required repeated dilations within a 14-day period over the course of four months until May 2007 (Fig. 3). Radiopaque markers became invisible 18 weeks after stent implantation and the oesophageal lumen at the former location of the stent, allowed passage of food. In May 2007, the patient started to be administered proton pump inhibitors (esomeprazole). The interval between dilations increased to four weeks. Positive results of the treatment (Fig. 4) improved the patient's health and normalisation of biochemical blood parameters convinced the parents and patient to continue therapy. In December 2007, the patient developed an oesophageal stricture at the level Th5–6. The second BD Stent (ELLA-CS) was placed into the lower part of the oesophagus. The patient tolerated the second implantation well, too. The oesophagus was checked using contrast fluoroscopy 24 hours, one week and then one month after stent placement.

Re-stenosis above the proximal stent end occurred 12 weeks after stent placement (Fig. 5). The lesion started to be treated by dilations performed within 4-week periods. Between dilations, the patient was able to eat solid food and visited the hospital at the moment when first symptoms of dysfagia forced him to eat only semi-solid food (dysphagia score 3). In June 2008, during fluoroscopic examination, previous axial hiatal hernia

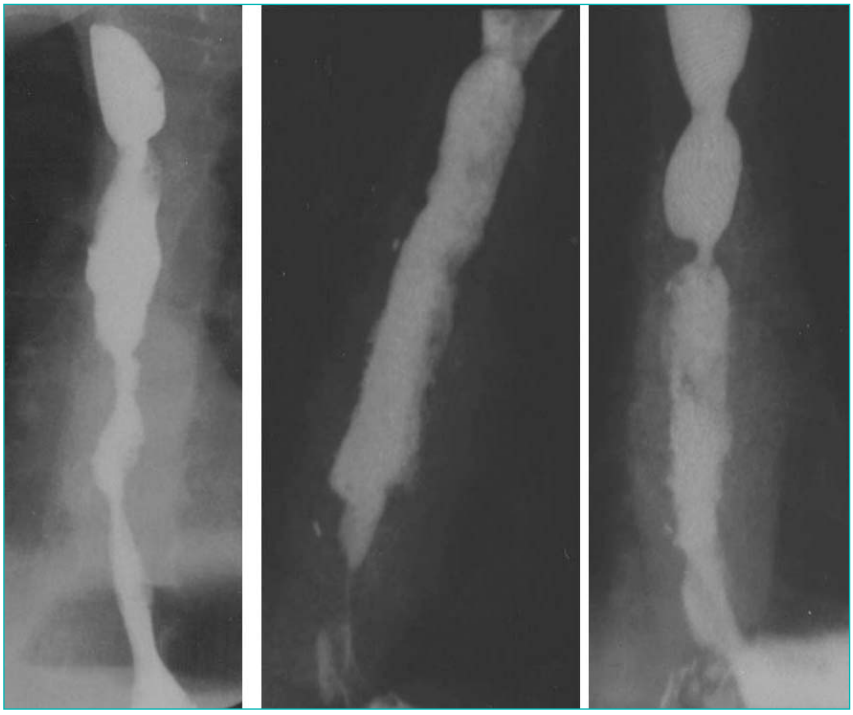

Fig. 1.
Corrosive oesophagitis with stenosis prior to implantation of the first stent (October 2006).
Fig. 2.
Fluoroscopic control five weeks after placement of the first stent (December 2006).
Fig. 3.
The oesophagus 11 weeks after placement of the first stent (February 2007).

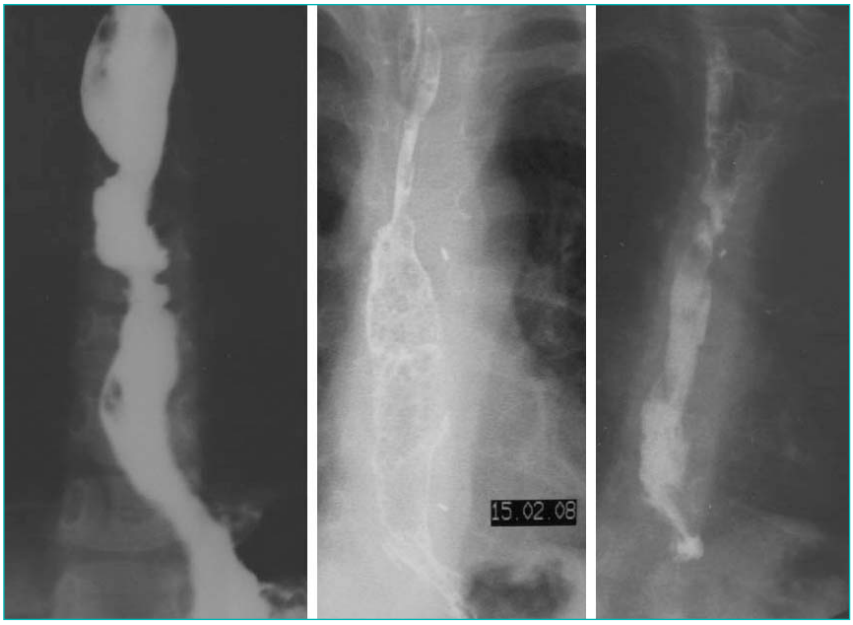

Fig. 4.
Symptomatic stenosis of the oesophagus prior to implantation of the second stent (October 2007).
Fig. 5.
Fluoroscopic control 12 weeks after placement of the second stent (February 2008).
Fig. 6.
The oesophagus 15 months after implantation of the last stent (February 2009).

and gastro-oesophageal reflux were not observed. Therapy with proton pump inhibitors was stopped due to non-compliance of the child's mother. In August 2008, severe stenosis occurred in the middle oesophagus narrowing it to a diameter of 2–3 mm. The situation was managed by balloon dilation. Esomeprazole administration was renewed and the interval between dilations increased to three weeks. Currently, the child is in a stage of moderate physical development. Two years after implantation of the first stent, the child's growth gain was 10 cm and weight gain 20 kg. The adequate therapy allowed the child to live a normal life, to visit a nursery and now a junior school. The patient hopes to recover and has confidence in himself. We intend to repeat stenting with a biodegradable oesophageal stent. Based on the experience, we want to use a stent longer than the previous one in order to place its ends at the healthy oesophageal mucosa and decrease the hyperplastic mucosal reaction (Fig. 6).

DISCUSSION

Management of caustic ingestion in children remains a difficult challenge, with the outcome ranging from an asymptomatic state to intractable esophageal strictures [2–5,8,11]. Moazam et al [6] reported 37 children ranging in age from 10 months to 5 years suffering from oesophageal burns. Twenty one (57%) of whom subsequently had oesophageal strictures of varying severity. In seven patients, limited oesophageal strictures were managed successfully by dilation, but 14 children with multiple strictures required eventual oesophageal replacement reportedly well-tolerated by patients. Other authors are very optimistic regarding oesophageal surgery in children with caustic stricture. Mutaf [7] presented the study involving 102 children with caustic oesophageal strictures requiring oesophagoplasty, 71 had a retrosternal colon transplant: two-stage oesophagocolostomy in 59 and one-stage cervical anastomosis in 12. In the retrosternal group, there were two cases of total transplant necrosis and three cases of terminal necrosis of the cervical end of the transplant; 12 patients developed anastomotic stenosis at the cervical end of the transplant. The incidence

of cervical anastomotic stenosis was 50% in the single-stage group compared to 10% in the two-stage group. The overall mortality was 3%. Twenty-six patients were followed up for 12 years: 21 (80%) had excellent results and 5 (20%) had residual problems. Mutaf states that associated morbidity remains significant since none of these patients has normal oesophageal motility after the substitution procedure. Therefore, every effort should be made to preserve the patient's own oesophagus.

The Pediatric-On-Line prepared by the American Academy of Pediatrics presents a website called "Esophageal Caustic Injury" at http://www.pediatriccareonline.org/pco/ub? [1]. The website summarises the principles and outcomes of oesophageal dilation as follows:

- Primary treatment with oesophageal dilation offers a satisfactory outcome for most otherwise healthy children with grade IIa injuries.
- Repeated dilation is rarely is successful for the most severe corrosive strictures; early surgical resection is associated with a better outcome.
- Oesophageal rupture is a potentially fatal complication of dilation and warrants immediate surgical repair. Incidence is 17% to 32%.
- Serial dilations are complicated by dysphagia between treatments, which may precipitate pulmonary aspiration.
- An adequate lumen is usually attained within six months to one year, with progressively longer intervals between dilations.
- Oesophageal replacement should be considered if dilation is ineffective beyond one year.

The first reports on stenting in children with caustic oesophageal strictures date back to the early 1990s. Coln & Chang [1] performed early implantation of silastic stents into a caustic lesion in children in order to avoid stricture formation. Wang et al [10] presented the study with 31 patients who underwent modified oesophageal intraluminal stenting through laparotomy 2–3 weeks after ingestion of the corrosive agent. The stent was removed without requiring anaesthesia after it had been in-situ for 4–6 months. Surprisingly the authors did not mention any mucosal reaction to long-term stent implantation as well as any complication related to the removal procedure. Five patients developed oesophageal stenosis from 2 to 3 months during follow-up.

Vandenplas et al [9] reported use of a BD Stent (ELLA-CS s.r.o., Czech Republic) in a 10-year-old boy with caustic stenosis in the mid oesophagus six weeks after ingestion of a corrosive agent. Although the patient remained symptom-free for four months, he developed a severe distal oesophageal stenosis over 4 cm about 10 months after the initial ingestion and six months after stent placement. Though the implantation brought temporary success, the author concluded that use of the BD Stent should be further evaluated as a first-choice intervention in patients developing a corrosive oesophageal stenosis.

Based on our clinical experience described above, we may conclude that the first BD stent managed the critical patient's condition and reduced the length of the stricture. Mucosal hypertrophy revealed by endoscopy does not require any intervention. We think the results of second stenting would have been more successful with a longer stent. The re-stenosis above the proximal and the absence of stenotic reaction below the distal stent end seem to suggest that the stent ends should be placed into healthy tissue. Recurrence of stenosis after discontinuing the treatment with proton pump inhibitors seems to prove that the patient should strictly continue the antireflux therapy during the entire treatment.

Based on experience with a child patient suffering from severe caustic oesophageal stenosis due to third-degree burns, we consider that bio-degradable stents might become a "golden therapeutic standard" improving the quality of the patient's life. However, further study and evaluation is needed in order to verify the positive clinical effect as to well as elaborate an appropriate therapeutic algorithm.

CONCLUSION

Although clinical experience is very limited, it seems that the BD Stent might represent a valuable therapeutic option in child patients with caustic stenosis intractable using standard dilation therapy.

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Correspondence to/
Adresa pro korespondenci:
Dr. Oksana V. Bychkova, MD
Children's Surgical Centre
1st Municipal Clinical Hospital
Prospekt Nezavisimosti 64
Minsk, 220030
Belarus
e-mail: oksanabychkova@mail.ru