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Use of covered self-expandable stents for benign colorectal disorders in children

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ABSTRACT

Purpose: There is a lack of experience with covered self-expandable stents for benign colorectal disorders in children.
Methods: Five children (4 M, 1F) with a median age of 5 years (range, 6 months–9 years) who underwent treatment with covered self-expandable plastic (SEPSs) or self-expandable metal stents (SEMSs) for a benign colorectal condition between April 2005 and November 2013 were recruited to this retrospective study. Etiologies included: anastomotic stricture with (n = 1) or without (n = 3) simultaneous enterocutaneous fistula, as well as an anastomotic leak associated with enterocutaneous fistula (n = 1). All children suffered from either Hirschsprung's disease (n = 3) or total colonic aganglionosis (Zuelzer–Wilson syndrome) (n = 2).
Results: Median duration of individual stent placement was 23 days (range, 1–87 days). In all cases up to five different stents were placed over time. At follow-up two patients were successfully treated without further intervention. In another patient the anastomotic stricture resolved fully, but a coexisting enterocutaneous fistula persisted. Overall, three patients did not improve completely following stenting and required definite surgery. Stent-related problems were noted in all cases. There was one perforation of the colon at stent insertion. Further complications consisted of stent dislocation (n = 4), obstruction (n = 1), formation of granulation tissue (n = 1), ulceration (n = 1) and discomfort (n = 3).
Conclusions: Covered self-expandable stents enrich the armamentarium of interventions for benign colorectal disorders in children including anastomotic strictures and intestinal leaks. A stent can be applied either as an emergency procedure (bridge to surgery) or as an adjuvant treatment further to endoscopy and dilatation. Postinterventional problems are frequent but there is a potential for temporary or definite improvement following stent insertion.

Insertion of colorectal self-expandable stents has been well established in adults. This technique is frequently used for palliation or as a bridge to surgery in patients with obstruction associated with malignant disease [1–3]. In addition, many authors reported on a more or less successful application of self-expandable stents in conditions such as diverticular disease, Crohn's disease, as well as ischemic, postsurgical anastomotic, and radiation-induced strictures [1,4–6]. However, there is a lack of sufficient long-term data and therefore stenting for benign colorectal disorders is not yet recommended as a first-line option [3].

Children may develop colorectal sequelae, including intestinal obstruction, perforation, anastomotic stricture and enterocutaneous fistula following surgery or from other underlying conditions such as inflammatory bowel disease and malignancy. However, physicians dealing with this group of patients have just begun to explore the use of self-expandable stents and up to now data are limited to a few case reports only [7–9].

We report our interdisciplinary experience with the use of covered self-expandable plastic (SEPSs) and self-expandable metal stents (SEMSs) for the treatment of benign colorectal conditions in children.

1. Material and methods

Five children (4 M, 1F) who underwent treatment with covered SEPSs or SEMSs for a benign colorectal condition between April 2005 and November 2013 were recruited to this retrospective study (Table 1). As part of an interdisciplinary approach patients were jointly managed from the Department of Pediatric Surgery and Central Interdisciplinary Endoscopy at our institution. The latter consists of general surgeons, pediatric surgeons, as well as adult and pediatric gastroenterologists. The overall median age at stent placement was 5 years (range, 6 months–9 years).

All children suffered from aganglionosis, either Hirschsprung's disease (n = 3) or total colonic aganglionosis (Zuelzer–Wilson syndrome) (n = 2).

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Table 1
Patients' characteristics and outcome.

No.	Sex	Age (yrs)	Underlying condition	Indication for stenting	Therapy prior to stenting[a]	Type of stent (size [mm]); stenting time	Complications	Outcome
1	m	9	HD	Anastomotic stricture 4 cm from anocutaneous line post Soave pull-through	Surgery (4×) including formation of loop-ileostomy, dilation (15×)	SEPS, 18–23/90; 19 days SEPS, 18–23/90; 12 days SEPS, 18–23/120; 52 days SEPS, 18–23/120; 8 days	Dislocation; ulceration; discomfort	Abdominoperineal resection and formation of end-ileostomy
2	f	4	HD	Anastomotic stricture 4 cm from anocutaneous line post TERPT	Formation of loop-ileostomy	SEPS, 16–20/120; 52 days SEPS, 18–23/90; 83 days SEPS, 18–23/90; 32 days SEPS, 21–25/90; 50 days	Perforation at stent insertion; dislocation,	Well at follow-up
3	m	5	TCA	Anastomotic stricture 4 cm from anocutaneous line and enterocutaneous fistula (initial pull-through procedure unknown)	none	SEPS, 18–23/90; 5 days SEPS; 18–23/90; 27 days	Dislocation; discomfort	Anastomotic stricture resolved, but fistula required resection
4	m	6	TCA	Anastomotic stricture 15 cm from anocutaneous line (initial pull-through procedure unknown)	none	SEPS, 16–20/120; 7 days SEMS, 16–20/120; 87 days SEMS pc, 18–23/120; 50 days SEMS, 24–26/120; 94 days SEMS pc, 18–23/120; 23 days[b]	Obstruction; formation of granulation tissue	Stricture resection
5	m	6/12	HD	Anastomotic leak and enterocutaneous fistula 2 cm from anocutaneous line post TERPT	none	SEMS, 10/80; 1 day SEMS pc, 16/40; 6 days SEMS, 18/120; 8 days[c] SEMS, 17–23/80; 2 days	Dislocation; discomfort	Well at follow-up

Abbreviations: m, male; f, female; yrs., years; HD, Hirschsprung's disease; TCA, total colonic aganglionosis (Zuelzer–Wilson syndrome); TERPT, transanal endorectal pull-through operation; SEMS, self-expandable metal stent; SEPS, self-expandable plastic stent; pc, partially covered.
[a] Primary pull-through operation and surgery related to the underlying condition not included.
[b] Placement of a second stent (18–23/120 mm) into the previous stent (telescoping) in order to achieve adequate overlap of the stenotic segment.
[c] Application of fibrin glue at the time of stenting.

1.1. Hirschsprung's disease

This cohort includes two boys (patient 1 and 5) and one girl (patient 2) who suffered from Hirschsprung's disease.

Patients 1 and 2 developed a distal anastomotic stricture after resection of the aganglionic portion of the colon: in one boy (patient 1) a stenosis 4 cm proximally of the anocutaneous line was diagnosed following a Soave pull-through operation. Despite numerous dilatations and surgery, including redo of the coloanal anastomosis and a diverting ileostomy, the stricture persisted. As an alternative option a covered self-expandable plastic stent (SEPS) was inserted. Another girl (patient 2) was referred to our center from another hospital with a distal stenosis following a transanal endorectal pull-through (TERPT). She did not improve after fashioning of a loop-ileostomy and a SEPS was placed subsequently.

A third infant (patient 5) suffered from an anastomotic leak and enterocutaneous fistula after a TERPT (Fig. 1). As part of an emergency operation during which an abdominal abscess was drained, a covered self-expandable metal stent (SEMS) was inserted endoscopically (Fig. 2). This maneuver was based on our experience with stenting for benign esophageal disorders in children [10] and in accordance with the parents who were extremely reluctant for the infant to undergo diverting stoma formation. In addition to stent therapy, an attempt to close the fistula by using fibrin glue was undertaken in the course of treatment.

1.2. Total colonic aganglionosis (Zuelzer–Wilson syndrome)

Two boys each (patients 3 and 4) with a total colonic aganglionosis developed an anastomotic stricture following surgery. They were transferred to our institution from abroad in a poor nutritional status. In either case the specific type of pull-through procedure performed could not be determined from the notes.

In patient 3 an additional enterocutaneous fistula adjacent from the anastomotic site was noted. The other boy (patient 4) suffered also from short bowel syndrome following multiple bowel resection and intestinal dilation.

In the course of treatment a self-expandable stent was inserted as a primary measure in both cases.

In all patients a variety of stents was placed (Table 1). From April 2005 to October 2013 SEPSs from different manufacturers (Willy Rüsch GmbH, Kernen, Germany; Polyflex® Esophageal Stent, Boston Scientific Corporation, Natick, MA, USA) were used. Most recently SEMSs that were coated with silicone and had radio-opaque markers were inserted (patients 4 and 5). Depending on patient's age, underlying condition and availability within our department we applied several types including esophageal, tracheal and biliary SEMSs from various companies (mandel + rupp, Erkrath, Germany; Boston Scientific Corporation, Natick, MA, USA; Mi.I. Tech, Seoul, South Korea). In the later cases (patients 4 and 5) partial covered stents

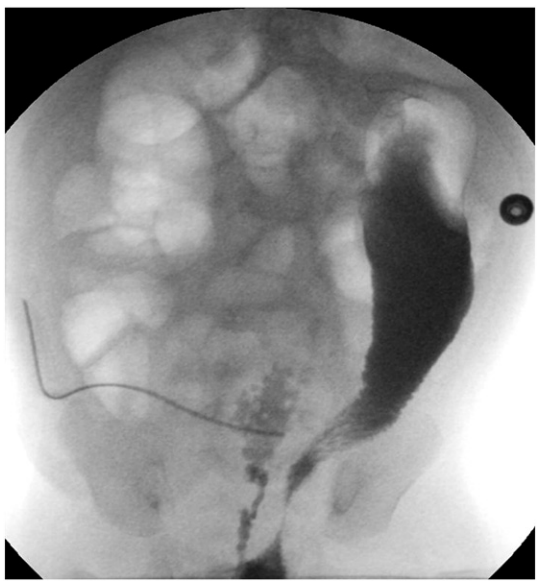

Fig. 1. X-ray showing an anastomotic leak and enterocutaneous fistula after a TERPT (patient 5).