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Use of Lumen-Apposing Metal Stents for Inflammatory Bowel Disease-Related Strictures at a Pediatric Center: A Case Series

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Lumen-apposing metal stents (LAMSs) including the fully covered, self-expanding metal AXIOS stent (Boston Scientific, Marlborough, MA) have been utilized for transgastric and transduodenal endoscopic drainage of pancreatic fluid collections in both adult and pediatric patients (1–3). The LAMS is barbell shaped with perpendicular flanges to help secure placement in tissue layers and prevent migration (1). The use of LAMSs has reportedly been used for management of selected luminal gastrointestinal strictures in adults (4–7). However, despite the frequent complication of stenosis particularly in Crohn's disease (8), literature review identified only one case detailing the use of a LAMS in an adult patient with inflammatory bowel disease (9). Further, no cases have been reported demonstrating the use of LAMSs in the management of gastrointestinal strictures of any origin in pediatric patients. In this series, we describe the use of the lumen-apposing, fully covered self-expanding metal AXIOS stent in 3 patients with inflammatory bowel disease. The use of AXIOS lumen apposing metal stents for luminal strictures is currently an unlabeled use of the commercial product.

METHODS

Three patients who had a LAM placed were identified by a group of 3 pediatric gastroenterologists who specialize in advanced therapeutic endoscopy for inclusion in this series. All 3 patients followed at our children's hospital gastroenterology clinic and share a diagnosis of inflammatory bowel disease. Indications for placement and outcomes varied (Table 1). In all cases, the LAMS utilized measured 15 mm in diameter by 10 mm in saddle length.

Received October 29, 2020; accepted December 24, 2020.

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The authors report no conflicts of interest.

All the authors fulfill the following criteria: substantial contributions to the conception or design of the work; or the acquisition, analysis, or interpretation of data for the work; drafting the work or revising it critically for important intellectual content; final approval of the version to be published; agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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JPGN Reports (2021) 2:2(e055)
ISSN: 2691-171X
DOI: 10.1097/PG9.0000000000000055

CASE PRESENTATIONS

Case 1

A 12-year-old female with a history of gastroschisis and atretic colon requiring colonic resection in the neonatal period, followed by small bowel obstruction requiring small bowel resection with Serial Transverse Enteroplasty procedure at 6 years old, and related short gut syndrome on fully enteral feeds presented with symptoms of nausea, early satiety, and nonspecific abdominal pain. She underwent colonoscopy and subsequent pathology review identified chronic active ileitis and epithelial ulceration with additional ulcers identified proximal and distal to the ileocolonic anastomosis. Together these findings were consistent with Crohn's-like pathologic changes and disease. The patient was subsequently treated with budesonide and later infliximab at a dose 5 mg/kg every 8 weeks without clinical improvement. Her serum infliximab concentration was monitored initially at maintenance (<1.0 μg/mL) and regularly thereafter. Her dose was increased stepwise to a dose of 10 mg/kg administered every 4 weeks to achieve a target serum infliximab concentration of greater than 5 μg/mL. Additionally, the patient did not develop detectable antibodies to infliximab at any point during her treatment. During this period, the patient was also started on monthly courses of metronidazole for management of small intestinal bacterial overgrowth. Repeat endoscopy 7 months from initial identification of ulcers identified an ileocolonic anastomotic stricture 5 mm in length which was dilated with the endoscope itself.

In the 15 months that followed, the patient's symptoms persisted despite ongoing medical management with metronidazole and infliximab at targeted concentration of greater than 5 μg/mL. In the week before repeat endoscopy, the patient had an elevated fecal calprotectin level of 508.7 μg/g and a serum infliximab concentration of 10.9 μg/mL. She underwent colonoscopy and the narrowed ileocolonic anastomosis was dilated with balloon dilation to a 12-mm luminal diameter. Following, a 15-mm diameter fully covered, LAMS was endoscopically placed at the stricture site (Fig. 1). Placement was assisted with on-table radiology.

Since the time of stent placement approximately 9 months ago, the patient has had resolution of nausea and early satiety, as well as markedly improved abdominal pain. The stent remains in place 9 months after placement and removal has been further delayed by familial preference in the setting of the ongoing COVID-19 pandemic.

Case 2

A 12-year-old male was initially evaluated for weight loss, decreased appetite, and elevated fecal calprotectin of 727.9 μg/g. He initially underwent esophagogastroduodenoscopy, but the endoscope was unable to be passed into the duodenum due to a stricture. He initially underwent esophagogastroduodenoscopy, but the endoscope was unable to be passed into the duodenum due to a stricture which measured approximately 3 cm in length and 4.8 mm in diameter as estimated relative to the endoscopic catheter diameter and its known

TABLE 1. Age and gender of patients who underwent lumen-apposing stent placement as well as indications, locations, outcome, and indwelling times

Patient age, gender	Indication	Location	Outcome	Indwelling time
12-year-old female (case 1)	Crohn's disease, anastomotic stricture, short gut syndrome	Ileocolonic	Symptom resolution, stent still in place	300 days*
12-year-old male (case 2)	Crohn's disease, refractory stricture with mucosal tear	Duodenal	Symptom resolution, stent removed successfully	21 days
21-year-old female (case 3)	Crohn's disease, anastomotic stricture	Ileocolonic	Symptom resolution, stent passed spontaneously	70 days

All stents utilized were 15 mm lumen diameter × 10mm saddle length.*Removal has only been delayed to the social restrictions in setting of COVID-19 pandemic.

dimensions. Following, the patient had an upper gastrointestinal series with small bowel follow through which confirmed the duodenal stricture presence. Magnetic resonance enterography (MRE) was performed and demonstrated inflammation involving the terminal ileum with bowel wall thickening and mucosal hyperenhancement concerning for IBD, though the duodenal stricture was not well visualized on MRE. Subsequent colonoscopy and pathology review identified active ileitis and chronic colitis with epithelioid granulomas consistent with Crohn's Disease. The patient was started on infliximab and nasogastric tube feeds with successful weight gain. Eight weeks following initiation of infliximab, the patient's serum infliximab concentration was 8.4 μg/mL and he had no detectable antibodies. Despite this, a repeat upper GI study 5 months following endoscopy demonstrated persistence of the duodenal stricture. The patient then underwent successful balloon dilation of the duodenal stricture from an initial diameter of 5–6 mm to 12 mm.

On repeat endoscopy 2 months following initial balloon dilation the patient was found to have persistence of the distal duodenal bulb stricture measuring 8 mm in diameter. The stricture was dilated with balloon dilation to a diameter of 12 mm. As part of our technique for persistent strictures, incisional therapy using a needle knife as a form stricturoplasty was performed. Following, a small mucosal defect with visible muscularis was noted, necessitating further intervention. The therapeutic endoscopy team felt that due to the location and angle that would be required to approach the defect, utilization of an over-the-scope clip alone would not be sufficient to achieve close approximation. Additionally, a self-expanding metal stent was considered, but there was concern that this type of stent would not only cover the mucosal defect, but the hepatopancreatic ampulla as well. A hemostatic clip was placed near the hepatopancreatic ampulla to mark its location. Following, a LAMS with a 15 mm diameter was placed across the stricture and mucosal defect with careful attention given to avoid covering the hepatopancreatic ampulla (Fig. 2). At time of placement, the hepatopancreatic ampulla was noted to be outside of the stented region and draining bile. Placement was assisted with on-table radiology. Following this intervention, the patient completed a 7-day course of prophylactic ciprofloxacin given the presence of the mucosal defect.

Three weeks later the stent was removed using rat tooth forceps. Upon removal, it was noted that the perforation defect had healed and the lumen diameter of 14 mm was maintained.

Two additional balloon dilations have been performed 2 and 8 months following the stent removal to maintain the luminal diameter at our goal of 12 mm. The patient has remained asymptomatic with further improved appetite and weight trajectory at time of his last endoscopy.

Case 3

A 21-year-old female with Crohn's disease and a history of ileocecal resection 1 year prior presented with worsening nausea, abdominal pain, and poor oral intake while on therapy with ustekinumab. Evaluation at this time included fecal calprotectin level of 35 μg/g, erythrocyte sedimentation rate of 19 mm/hr and C-reactive protein level <0.3 mg/dL. At this time, serum ustekinumab concentration was drawn at 6 weeks following induction and found to be above targeted concentration of 6.0 μg/mL with no detectable antibodies. However, MRE was performed and demonstrated mild inflammatory changes involving a short segment of ascending colon and inflammatory changes involving 7 cm of distal ileum adjacent to the ileocolonic anastomosis. A short segment of persistent

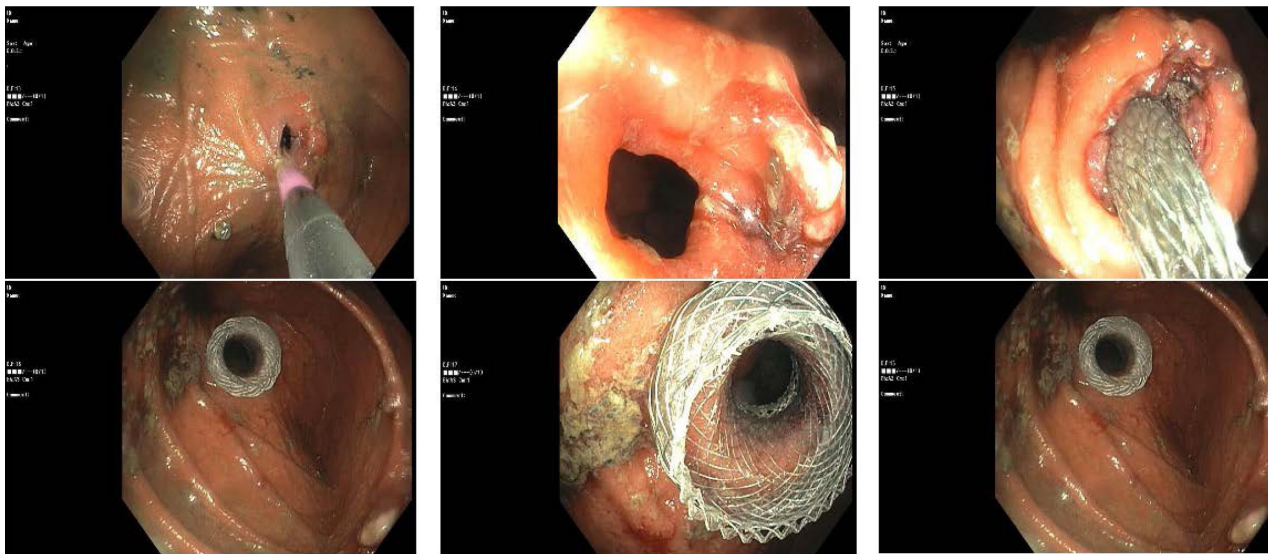

FIGURE 1. Gallery of select images from case 1 at time of fully covered, lumen-apposing metal stent placement.

luminal narrowing measuring 4–5 mm was also visualized on MRE at the level of the ileocolonic anastomosis. Given the inflammatory changes, it was decided to trial medical management with methylprednisolone and initiation of adalimumab. Despite this change in medical management, the patient's symptoms persisted in the ensuing 3 months and she then underwent upper GI study with small bowel follow through which redemonstrated this short stricture at her ileocecal anastomosis. Colonoscopy was performed which identified the ileocolonic anastomosis site with a short, fixed stricture measuring approximately 3–4 mm in diameter and 5 mm in length as approximated relative to the scope itself. This stricture was successfully dilated with a balloon to 12 mm and further to 15 mm on repeat colonoscopy 1 month later.

Following repeat balloon dilation, the patient's symptoms of postprandial pain and nausea had not resolved. Six weeks later, she underwent placement of a 15 mm diameter LAMS via colonoscopy. The stent was placed over a wire support and guided by fluoroscopy (Fig. 3).

Approximately 3 months later, the patient underwent colonoscopy to remove the stent. However, the stent was not visualized and was presumed to have passed spontaneously. The luminal diameter maintained an open position of 15 mm. At the patient's subsequent clinic visit 7 months later, symptoms of abdominal pain and nausea had not recurred. Following this last visit, the patient transitioned care to Adult Gastroenterology.

DISCUSSION

In this single-center series of 3 cases, LAMS s were successfully placed in IBD patients for various strictures and related

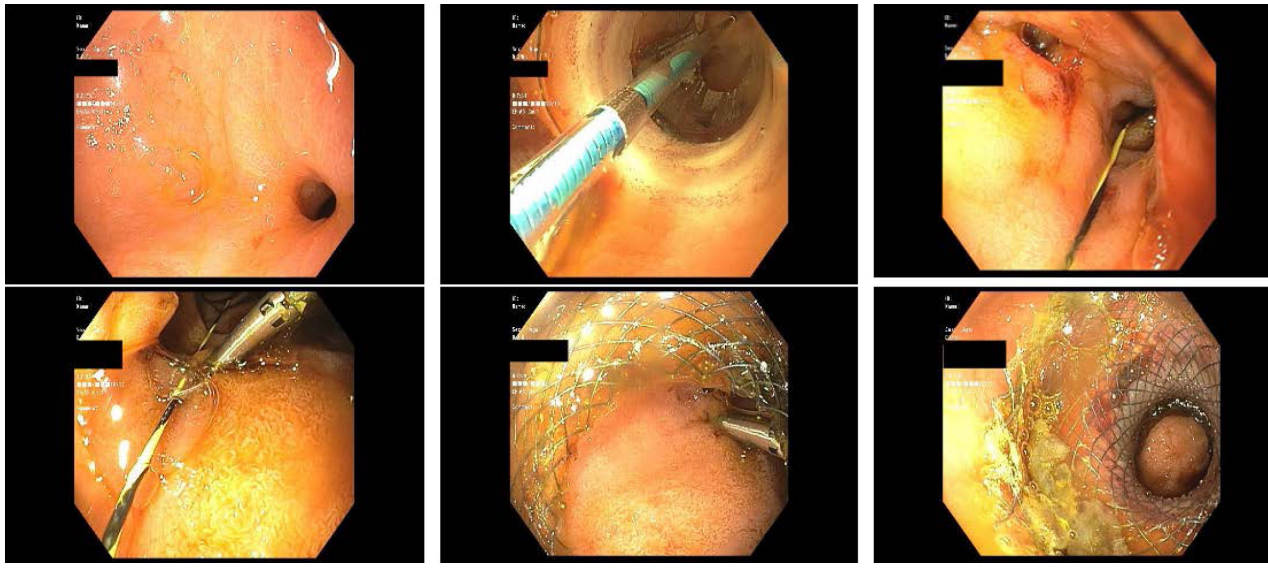

FIGURE 2. Gallery of select images from case 2 at time of lumen-apposing metal stent placement.

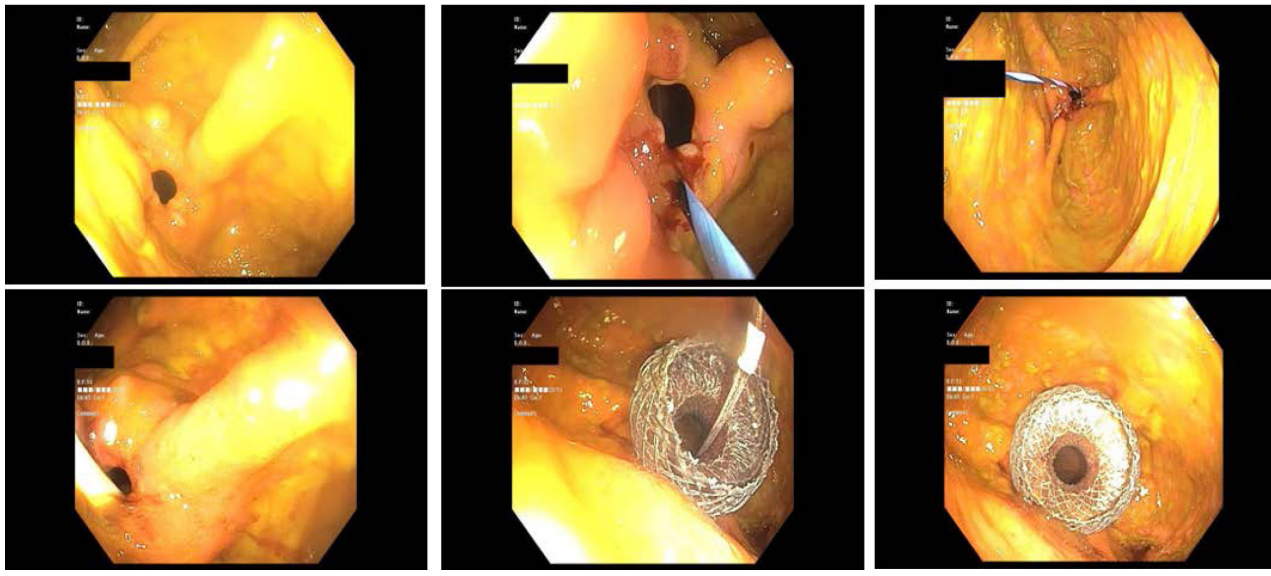

FIGURE 3. Gallery of select images from case 3 at time of fully covered, lumen-apposing metal stent placement.

complications by gastroenterologists with experience in therapeutic endoscopic procedures. In our series, the LAMSs were utilized as a next-step therapy to balloon dilations and when complications were encountered. In all instances, patients reported clinical improvement in their symptoms following LAMS placement. In 1 of the 2 cases that has been reevaluated by endoscopy following placement, the luminal diameter was maintained at 15 mm. There was noted variability in stent indwelling time with the stent passing spontaneously in 1 case. Our team does not routinely recommend fixation of LAMSs unless the patient becomes symptomatic following device migration or in the event that persistence or recurrence of the luminal stricture necessitates the placement of a new stent.

This case series suggests there may be a role for wider utilization of fully covered, LAMSs such as the AXIOS stent in select pediatric and young adult cases of refractory or complicated luminal strictures related to inflammatory bowel disease. Our review illustrates that placement of LAMSs is technically possible in strictures both in the small and large intestines. Our successful experiences align with similarly reviewed cases in adult centers (7,9). Given the learning curve for placement of stents, it is our opinion that these stents should be reserved mainly for gastroenterologists who have significant prior experience in therapeutic endoscopic modalities including luminal stent placement. We prefer a stent indwelling time of 4–6 weeks and reevaluation of stent location with abdominal radiographs if needed. The manufacturer reports the stent is MRI-conditional as determined by nonclinical testing. Future prospective studies with larger samples are needed to substantiate our success, define risks and safety considerations, and to compare findings with conventional interventions such as balloon dilation.

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