RESEARCH

Double balloon enteroscopy in children: a case series highlighting risks, and a review of the literature

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ABSTRACT

Aim Double balloon enteroscopy (DBE) is a relatively new endoscopic technique that allows endoscopic access into the small intestine. It has been studied in adults, but the literature remains scarce in the paediatric age group.

Methods We retrospectively assessed our experience with DBE in children.

Results We describe four procedures performed on three patients. The diagnostic yield was high (100%) with one major complication (perforation) encountered. A review of the benefits and risks in this cohort, and the available literature is included.

Conclusions DBE is a promising endoscopic modality in children, but some aspects remain unanswered. There is a great need for a prospective study to better assess benefit and risk in this population.

INTRODUCTION

Double balloon enteroscopy (DBE) is a new technique that allows endoscopic access into the small intestine.^{1 2} Previously used non-surgical modalities did not allow endoscopic examination of the entire small intestinal length or performance of interventions, such as obtaining biopsies, polyp removal or treatment of bleeding lesions. Since its introduction in 2003, DBE has been studied in adults, but the literature remains scarce in the paediatric age group. We present our experience with DBE, describing four procedures performed on three patients at our facility.

METHODS

This was a retrospective chart review of DBE procedures in paediatric patients at the University of Iowa from January 2010 to May 2011. Patients were identified through a procedure database.

Inclusion criteria included age <18 years at time of the procedure. Follow-up time was defined as the time from the initial DBE procedure until the last documented clinic or phone encounter. The institutional review board of University of Iowa approved the study.

The equipment included a double-balloon therapeutic enteroscope (Fujinon EN-450T5, Fujinon Inc, Saitama, Japan) with a length of 200 cm, outer diameter of 9.4 mm, forceps channel diameter of 2.8 mm, and a flexible overtube with an outer diameter of 13.2 mm. Each procedure was performed by the same two physicians, a paediatric and an adult gastroenterologist. The technique involved a series of motions with enteroscope and overtube insertion and balloon inflation and deflation to allow advancement into the small bowel. Assessment of the length of small bowel visualised was carried out as previously described by May *et al.*³ No fluoroscopy was involved. All procedures were performed under general anaesthesia with procedural time limited to less than 2 h. Bowel preparation used was similar to our standard colonoscopy bowel preparation with 4 days of polyethylene glycol.⁴ The type of DBE procedure (antegrade vs retrograde) was chosen based on the available clinical information including imaging or capsule endoscopy results.

RESULTS

We identified three cases (all males) that underwent four procedures. The cohort demographics, procedure characteristics and endoscopic findings are summarised in table 1. The following are the detailed description of each case:

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Table 1 Cohort summary

Case	1	1	2	3
Procedure	1	2	3	4
Age (yrs)	11	11	7	13
Weight (kg)	30	28.2	36.3	43.9
Procedure type	Antegrade	Retrograde	Antegrade	Antegrade
Indication	GI bleed	GI bleed	Abdominal pain	Intestinal polyps
Insertion depth (cm)	175	70	275	90
Findings	+	+	+	+
Complications	–	–	–	+

yrs, years; antegrade refers to an oral approach; retrograde refers to an anal approach; +, present; –, absent; GI, gastrointestinal.

Case 1

An 11-year-old boy with known short-bowel syndrome on full enteral nutrition was admitted to the intensive care unit with new onset rectal bleeding and significant anaemia (hemoglobin 5 g/dl) requiring packed red blood cell transfusions. His workup included a negative esophagogastroduodenoscopy (EGD) and colonoscopy. A DBE was performed to assess for a bleeding source within the small bowel. His past medical history included gastroschisis with bowel ischaemia and necrosis with subsequent short-bowel syndrome. He had multiple surgeries, including a Bianchi bowel-lengthening procedure at 6 months of age, and a recent laparotomy at 10 years of age for small bowel obstruction. The recent surgery revealed an internal jejunal volvulus around an adhesive band that was lysed, in addition to takedown of Bianchi anastomosis site (for concerns about obstruction), resection of 8 cm of terminal ileum, and a serial transverse enteroplasty (STEP) procedure on the existing dilated small bowel.

An antegrade DBE was performed revealing an anastomotic small bowel site ulceration, with a visible adjacent surgical clip, likely the site of his STEP procedure. The area was marked with India ink and a clip. Around 10 cm distally, another area of ulceration was noted and marked with India ink. Despite the use of a stiffening wire and application of abdominal pressure, further advancement was not possible. An estimated total of 175 cm of the small bowel was examined. The enteroscope was withdrawn without complications.

The patient continued to have intermittent rectal bleeding and underwent exploratory laparotomy (15 days later). A 45 cm small bowel segment was resected, including the ulcerated area guided by the tattooed mark placed during DBE. Intraoperative push endoscopy through the remainder of the distal small bowel into the colon did not show other potential bleeding sources. He was discharged home but presented with rectal bleeding 4 months later. A tagged red blood cell (RBC) scan was negative.

A retrograde DBE was offered. The enteroscope was advanced through the colon into the small bowel. Enteroenteric anastomotic ulcers were noted 5 cm proximal to the ileocolonic anastomosis without active bleeding. The enteroscope was advanced another 70 cm into the proximal small bowel without other abnormalities noted. The ulcerated area was treated using gold probe coagulation (settings of 30 Watts, effect 2) (figure 1). The enteroscope was withdrawn without complications. On follow-up 1 year later, the patient remained symptom-free without further bleeding episodes.

Case 2

A 7-year-old boy presented with episodic abdominal pain and vomiting of 2 months duration, along with decreased appetite and a 4–5 kg weight loss. His family recalled a painful maculopapular rash on his lower extremities 2 weeks prior, which was not present on evaluation. His exam showed periumbilical and left lower quadrant tenderness. Stool was haemoccult positive, with normal other labs including, complete blood count and liver function tests. An abdominal CT scan revealed segmental small bowel wall mesenteric thickening without obstruction. Upper gastrointestinal series with small bowel follow-through revealed two suspected strictures in the proximal small bowel with a normal terminal ileum. EGD showed gastric and duodenal bulb erythema. Colonoscopy was unremarkable. Biopsies were normal except for mild chronic gastritis. Other workup included negative serology 7 (IBD7) panel (Prometheus Laboratories, San Diego, California). He was started on oral controlled-release mesalamine formulation and corticosteroids for presumed Crohn's disease. The patient returned 2 weeks later with symptom improvement. He had no prior surgeries.

An antegrade DBE was performed 4 weeks after initial presentation to better assess for small bowel

Figure 1 Enteroenteric anastomotic ulcer after treatment with gold probe coagulation.

Crohn's disease. The enteroscope was advanced approximately 275 cm into the small bowel. The extent of the examination was marked with India ink. Two ulcers were noted in the distal small bowel. One was towards the distal extent of the examination and could not be biopsied. The second was ~100 cm proximally (figure 2A) and cold biopsies were obtained. The enteroscope was withdrawn without complications. Histopathology was non-specific showing an oedematous submucosa with few scattered neutrophils and haemorrhages at the tips of the villi. The patient returned 3 weeks later with abdominal pain, haematemesis and hematochezia. Repeat EGD and colonoscopy were normal. Following a third similar presentation, an exploratory laparotomy was performed with small bowel resection, including the tattooed area (figure 2B). Histopathology of the resected segment showed expansion of the serosal surface and few areas of haemorrhage at the epithelial surface with no signs of inflammation or vasculitis. The patient was tapered off corticosteroids. Ten months later, a palpable purpuric rash erupted mostly affecting the lower extremities. A skin biopsy revealed leukocytoclastic vasculitis suggestive of Henoch

Schonlein purpura (HSP). Follow-up 10 months after surgery showed that the patient had recurrent HSP episodes including abdominal pain, managed with steroids.

Case 3

A 13-year-old boy with known Peutz-Jeghers syndrome (PJS) underwent screening capsule endoscopy to evaluate for small bowel polyps showing three large polyps, likely in the ileum. His medical history included a PJS diagnosis at 3 years of age. He had several surgeries, including surgical reduction of a polyp-induced small bowel intussusception, polyp removal, and resection of non-viable small bowel at 4 years of age, and subsequent surgical removal of a large polyp at the ileocecal valve at 6 years of age. He had surveillance EGD and colonoscopy with multiple polyps removal every 2 years.

An antegrade DBE was performed to remove the identified small bowel polyps. A 3 cm diameter polyp (figure 3A) was noted in the distal small bowel. The polyp could not be grasped entirely and was removed piecemeal (figure 3B) using a polypectomy snare (settings 30 Watts, effect 3, coagulation 4). Another smaller polyp was noted distally and removed in one piece using the same method. No air bubbling or signs of perforation were noted. Around 90 cm of small bowel length was examined, and further advancement was not possible despite use of a stiffening wire and application of abdominal pressure. The patient seemed to tolerate the procedure well except for minor abdominal discomfort, and was discharged home. He returned the next day with severe abdominal pain, and abdominal films showing free intraperitoneal air (figure 3C). An exploratory laparotomy identified a small perforation with resection of 8 cm small bowel segment. The patient was discharged 1 week later. He was doing well on follow-up 5 months later.

DISCUSSION

The diagnosis and treatment of small bowel pathology has been historically difficult. Capsule endoscopy provides visualisation of the entire small bowel, but does not allow specimen acquisition or procedural interventions. In the adult population, DBE has been shown to be instrumental as a diagnostic and therapeutic modality for management of small bowel pathology. The main indications in adults include obscure gastrointestinal (GI) bleeding, histologic confirmation or resection of small bowel lesions, stricture dilation, placement of preoperative marking of pathologic findings, and foreign body removal.⁵⁻⁷ The diagnostic yield is high, in the 72%–80% range^{3 8} with a low complication rate of 0.8% for diagnostic procedures, and 4.3% for therapeutic procedures.⁹

The limited studies on DBE in children describe it as safe and clinically useful, but these are mostly retrospective and include relatively small numbers.

Figure 2 (A) Small bowel ulcer noted during enteroscopy. (B) Tattooed area identified during exploratory laparotomy (Courtesy of Dr A Shaaban, Division of Pediatric Surgery, University of Iowa).

Figure 3 (A) Large polyp noted in the distal small bowel in a patient with Peutz-Jeghers syndrome. (B) Same polyp following piecemeal removal. (C) Abdominal film showing free intraperitoneal air from a small bowel perforation.

Indications include GI bleeding, abdominal pain, chronic diarrhoea, suspected small bowel pathology, stricture or polyp, and assessment for biliary strictures after liver transplantation and Roux-en-Y hepaticojejunostomy.^{10–12} Diagnostic yield ranges between 46% and 88%.^{10–13} Postprocedural sore throat is frequently reported.^{10–11–13} While some series have had no serious complications,^{10–13} others reported aspiration,¹¹ postpolypectomy bleeding¹² and small bowel perforation.¹⁴ One patient developed an intra-abdominal abscess following laparoscopic-assisted DBE (with small bowel polyp removal).¹⁵ Failure to intubate the ileocecal valve during retrograde DBE has been reported.^{13–14} As of now, it is not possible to correlate patient age with complication rates, as several studies did not include the age of patients who developed specific complications. A summary of available paediatric studies is presented in table 2. By comparison, the incidence of complications during diagnostic

upper endoscopy and colonoscopy in children is less than 1%.^{16–17}

In our series, the overall yield was high, with abnormal endoscopic findings noted in all cases. Our series highlights several aspects pertaining to benefits and risks of DBE in children. The procedure proved beneficial in our first case, with identification of a surgical complication (anastomotic ulcer following a STEP procedure) and guidance of subsequent surgical intervention through injection marking. With symptom recurrence, the same patient benefited after endoscopic therapeutic intervention for an intermittently bleeding ulcer. In our second case, endoscopic abnormalities were noted, but the benefit was limited, as a definitive diagnosis could not be made. Biopsy findings were non-specific, possibly influenced by prior initiation of systemic steroids. A possible benefit was the lack of evidence to support a diagnosis of Crohn's disease. The diagnosis remained unknown even after surgical

Table 2 Summary of published reports

Reference number, publication year	Number of patients	Number of procedures	Age range (yrs)	Yield (%)	Major complications
#, ¹⁰ 2007	26	30	4–29	23/26 (88)	None
#, ¹¹ 2009	31	33	3–14	25/31 (81)	1 aspiration
#, ¹² 2010	48	92	4–18	31/48 (65)	1 post polypectomy bleed
#, ¹³ 2010	11	13	8–20	6/13 (46)	None
#, ¹⁵ 2010	14	21	8–16	11/14 (78)	1 abdominal abscess (laparoscopic-assisted)

#, reference number; yrs, years.

exploration and resection of the marked small bowel segment. The patient likely had HSP with small bowel involvement that became apparent with the later onset of cutaneous manifestations. Our third case developed a significant complication (perforation) requiring surgical intervention. Although the risk could not have been reduced with the presence of a large polyp, earlier recognition of a perforation could have been achieved. Following therapeutic DBE, radiologic abdominal imaging prior to discharge, or an overnight observation should be considered. Although less invasive compared with intraoperative enteroscopy, DBE may be limited, or not feasible, in patients with prior abdominal surgeries due to adhesions or dilated small bowel segments that can hinder balloon anchoring. This may have been a factor in the limited insertion depth encountered in our last case.

The technique of DBE was described by Yamamoto in 2001.¹ In the paediatric population, the first report was published in 2007;¹⁰ however, the series by Nishimura *et al* clearly described the use of DBE in children starting May 2003.¹² This advanced endoscopic modality can be beneficial in the identification and treatment of small bowel abnormalities. Complications are possible and can be life threatening, including perforation. Similar to the experience with adults, complication rate is likely dependent on procedure type (therapeutic vs diagnostic), but other risk factors (patient size, prior abdominal surgery) may play a role. Several barriers to the adoption of DBE by paediatric gastroenterologists including lack of appropriately sized equipment for younger patients, fewer indications in this population, cost and training. Due to the several limitations stated, DBE has not been adopted widely as the standard of care for specific disorders/disease processes. Many aspects of DBE in children remain unanswered, including optimal bowel preparation, sedation type, and postprocedural observation. Many of these questions would be best addressed in a large multicentre prospective study.

Key messages

This study illustrates the high yield of DBE in the evaluation of small bowel disease in children but it also highlights the potential limitations and risks. The study emphasizes the need for a prospective trial to investigate factors that can help treating physicians balance risk and benefit when considering this endoscopic modality.

Contributors All authors have contributed significantly to the manuscript and are in agreement with its content.

Competing interests None.

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