

Successful Laparoscopic Treatment of Hemorrhage from Ileal Duplication Cyst in a 10-Year-Old Saudi Boy

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Abstract

Lower gastrointestinal hemorrhage often produces diagnostic difficulty in children. Endoscopy, radionuclide scans, computed tomography/magnetic resonance angiography, and conventional angiography have been the usual armamentarium for locating the site of bleeding. In recent times, laparoscopy has offered an effective tool for diagnosing various intra-abdominal, especially extraluminal, pathologies elusive to other modalities of diagnosis. In this paper, we present a case of massive hemorrhage from an ileal duplication cyst in a 10-year-old Saudi boy diagnosed and treated by laparoscopy. We believe this is the first such reported case from the Kingdom of Saudi Arabia.

Introduction

LOWER GASTROINTESTINAL (GI) HEMORRHAGE due to its obscurity and episodic nature is always a challenge to the surgeons and physicians. Children and infants have different causes, as compared to the adults. Investigative modalities, such as endoscopy (colonoscopy, upper GI endoscopy), radionuclide scans (Meckel's scan, red blood cell [RBC] pool scan), small-bowel enema, computed tomography/magnetic resonance (CT/MR) angiography, and conventional angiography have been used with various degrees of yield to locate the site of hemorrhage, depending on the policy and availability in a given setup. In certain cases of an obscure lower GI bleeding laparotomy, the transillumination of the bowel, enterotomy, and intraoperative endoscopy of the small bowel have been advocated. In the recent era of minimal access surgery, laparoscopy offers a valuable tool in such cases. We describe in this paper a case of massive hemorrhage from an ileal duplication cyst in a 10-year-old boy diagnosed and treated by laparoscopy.

Case Report

A 10-year-old Saudi boy presented to the pediatric emergency room with complaints of massive bleeding per the rectum of 12 hours in duration. There was no associated abdominal pain, vomiting, fever, or preceding history of diarrhea. The parents denied a history of excessive bleeding from injury sites, easy bruising, or bleeding from the gums. There was no history of similar complaints in the family. He

gave a history of two episodes of painless bleeding per the rectum in the last 2 years, which subsided spontaneously without any medical intervention on both occasions. There was no history of blood transfusion during any such episodes in the past. Upon evaluation, the boy was found to be pale and tachycardic with hypotension (blood pressure, 88/60 mm Hg). The abdomen was soft, lax, flat, and nontender. The rectal examination revealed fresh and clotted blood without any palpable mass or polyps. His investigations showed hemoglobin of 6 g/dL, normal counts and prothrombin, and activated partial thromboplastin time. The ultrasound evaluation of the abdomen was essentially normal. He was resuscitated with a packed cells transfusion and intravenous fluids. An upper GI endoscopy and colonoscopy after a purged bowel preparation failed to locate the site of bleeding. Due to the recurrent massive, obscure nature of the lower GI bleeding, he was, after discussing with the parents, subjected to diagnostic laparoscopy under general anesthesia. Laparoscopy was performed through a 5-mm umbilical port assisted by two other 5-mm ports, one at the right-upper quadrant (midclavicular line) and the other at the suprapubic site (Fig. 1). The ileocecal junction (IC) was identified and the ileum was traced proximally for a possible Meckel's diverticulum. About 70 cm proximal to the IC junction, a 4×5 cm² cystic lesion at the mesenteric side of the ileum was detected (Fig. 2). There were lobulations and congestion on its surface. Antimesenteric surface of the bowel was normal, and there was no prominent vessel suggestive of the vitelline artery supplying the cyst. A diagnosis of an ileal duplication cyst was made and the rest of the bowel was explored to rule

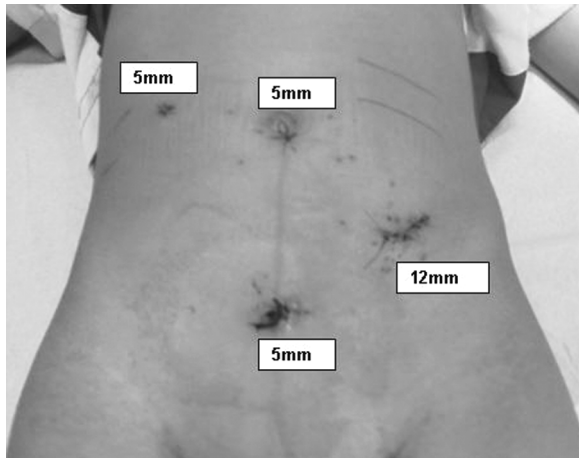


FIG. 1. Position of the umbilical (5-mm), right hypochondrium (5-mm), suprapubic (5-mm), and left iliac fossa (12-mm) ports.

out any other synchronous lesions. A resection of a segment of ileum with the duplication cyst and side-to-side ileoileal anastomosis was done by the use of an Endo-GIA stapler (Ethicon, Cincinnati, OH; Endoscopic Articulating Linear Cutter, size 45 mm, introduced through an additional 12-mm left iliac fossa port) and intracorporal suturing. The division of mesenteric vessels and duplication cyst was aided by a 5-mm Harmonic Scalpel (Ethicon, Cincinnati, OH). Only the 12-mm port site had fascial closure, and the three 5-mm ports had skin closure only. The total operating time was 90 minutes, without any intraoperative complications. The boy recovered smoothly, was allowed orally on day 3 and was discharged home on the postoperative day 4. At the subsequent follow-up, his wounds had healed completely without any evidence of port-site hernia and he had no further bleeding. Histopathology was consistent with a communicating ileal duplication cyst with heterotopic gastric mucosa and ulcerations (Fig. 2).

Discussion

Massive lower GI bleeding in children is alarming and anxiety provoking to the parents as well as the physicians. Often, the course is self-limiting and episodic, as in this index case. The causes are usually straightforward in children and include polypoid disorders, Meckel's diverticulum, bleeding disorders, or Henoch Schönlein purpura. These are readily detectable by a careful history, clinical evaluation, bleeding profile, GI endoscopy, and a Meckel's scan. Sometimes, sources of bleeding are obscure and are caused by alimentary duplications, hemangiomas, arteriovenous malformations, lymphonodular hyperplasia, Dieulafoy's ulcer, and vasculitis enterocolitis.

Alimentary duplications are cystic or tubular structures attached to the mesenteric side of the gut, often sharing a common muscle wall and vascular supply. It is lined by intestinal mucosa, which may be different from the area wherefrom it is arising (i.e., differentiates from other mesenteric cysts).¹ Meckel's diverticulum is differentiated by its blood supply by a separate vitelline artery (branch of the superior mesenteric artery) and its antimesenteric location.² Bleeding

from the duplication cyst due to ectopic gastric or pancreatic mucosa is rarely massive and is seen in around 35% of cases³ (as in the present case). Bleeding has also been reported to occur due to intussusception and pressure necrosis of the adjoining intestinal wall.⁴

Although quite popular, the role of scintigraphy by Tc-99 pertechnetate to delineate ectopic gastric mucosa in Meckel's diverticulum (a Meckel's scan) or duplication cyst in the setting of massive lower GI bleeding (LGIB) is questioned by many.⁵⁻⁷ The sensitivity of the Meckel's scan falls from 80 to below 60% in cases of massive bleeding and at a hemoglobin level below 11 g/dL.⁵ Most of the symptomatic patients who present to the hospital usually have low hemoglobin, thus negating the usefulness of scintigraphy. Sensitivity of scintigraphy is further low, especially in cases of duplication cysts with a small amount of functioning gastric mucosa, nonfunctioning/necrosed mucosa, and dilution of secreted isotope as a result of hemorrhage or rapid intestinal transit due to the irritating presence of blood within the lumen or the presence of activity in an overlapping organ (e.g., urinary bladder).⁸⁻¹⁰

Some advanced, sophisticated diagnostic modalities, such as the RBC pool scan, CT/MR angiography, enteroclysis, and so on, have been employed in adults (causes are different from children) in the evaluation of LGIB with suboptimal sensitivity and specificity.¹¹ Besides the lack of access to such modalities in emergency situations, the technical difficulties, lack of standardization of procedures, need of anesthesia in children, and difficulty and morbidity associated with some of the procedures such as conventional angiography have made their role redundant or secondary in children with lower GI bleeding.

In contrast, laparoscopy has evolved in recent decades as a useful, accurate tool in the evaluation and treatment of various intra-abdominal pathologies.¹²⁻¹⁴ Hence, it has been our policy also, like others, over the past few years, to use laparoscopy as an investigative and therapeutic modality for children presenting with small-bowel bleeding.¹⁵⁻¹⁷ Although we perform the colonoscopy and upper GI endoscopy whenever possible, prior to laparoscopy in

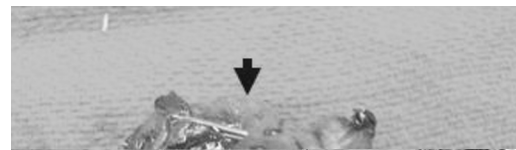


FIG. 2. Resected ileal duplication showing communication and ectopic gastric mucosa (open arrow). Bold arrow shows the antimesenteric border of the ileum.

our cases, some researchers have recommended diagnostic laparoscopy ahead of bidirectional endoscopy in the evaluation of LGIB, as air insufflation during the procedure might hinder a successful laparoscopic evaluation under the same general anesthesia.¹⁵

Resection of bleeding Meckel's diverticulum has been accomplished by laparoscopy-assisted extracorporeal bowel resection^{14–18} or intracorporeal suturing and stapling techniques.¹⁹ However, there are anecdotal reports of the complete laparoscopic treatment of alimentary duplications²⁰ in children in the literature, so far. Laparoscopic segmental resection of the ileum (along with the duplication cyst) with intracorporeal suturing and the application of Endo-GIA staplers was performed safely in our case and the use of the Harmonic Scalpel aided in the division of mesenteric vessels supplying the bleeding duplication cyst.

Conclusion

The source of GI bleeding in children may become obscure to standard endoscopic modalities due to conditions such as intestinal duplication anomalies. Laparoscopy is an ideal modality to identify such obscure sources and also allows definitive therapy at the time of evaluation. Modern laparoscopic devices such as the Harmonic Scalpel, and Endo-GIA staplers enable the experienced laparoscopic surgeon to perform intracorporeal intestinal resection and anastomosis with ease and rapidity even in emergency situations, such as massive lower GI bleeding.

Disclosure Statement

No competing financial interests exist.

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