



Case Report

Perirectal Abscess and Fistula Secondary to a Fecalith in Rectal Pouch Occurring 51 Years After Duhamel Operation for Hirschsprung's Disease: Minimally Invasive Treatment

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Abstract

Background: Duhamel pull-through procedure is one of the techniques performed in Hirschsprung disease cases. Fecalith formation secondary to a rectal blind pouch or remnant colorectal septum is a common complication after procedure. Fecalith could lead to the area of ischemic colorectal pressure ulcers, causing localized ischemic necrosis and colorectal perforation. Endoscopic Vacuum Therapy (EVT) may be minimally invasive management technique of GI defects.

Case Presentation: We report a 51-year-old man, who had undergone a Duhamel operation for Hirschsprung's disease at one year of age, presenting with a faecalith in the blind rectal pouch, which presumably eroded into the rectum and developed the mesorectal abscess and fistula to lesser pelvis. EVT was used to treat pelvirectal fistula and defect in the rectum wall. Pelvic MRI was performed two months after discharge-no fistula or rectal pouch wall defects were shown.

Conclusions: The EVT treatment of perirectal abscess and fistula could be one of the most promising applications of vacuum-assisted closure in coloproctology. Treatment strategy should be adopted individually.

Keywords: Duhamel operation; Endoscopic vacuum therapy; Hirschsprung disease

Introduction

Hirschsprung Disease (HSCR) is the congenital absence of enteric ganglion cells (Aganglionosis) in the distal intestinal tract, with

more subtle proximal alterations at the Transition Zone (TZ) between normal and abnormal bowel. These neuroanatomic alterations correlate with intestinal spasticity and chronic bowel obstruction [1]. Adult cases of this condition are relatively rare, mostly found in males (male:female ratio of 4:1) and associated with trisomy 21 or other genetic syndromes [2]. Surgical intervention with resection of aganglionic bowel is the principal

treatment for Hirschsprung disease. The operation is usually performed early in life. Duhamel pull-through procedure is one of the techniques performed in HSCR cases [3]. The Duhamel technique involves a retrorectal transanal pull-through method [4] (Figure 1). The rectum's posterior wall and the pulled-through colon's anterior wall are opposed by a crushing clamp resulting in a wide anastomosis. The aganglionic rectum is left connected to a segment of the ganglionic colon (side-to-side) as a pouch reservoir [5]. The original Duhamel procedure has the benefit of less pelvic dissection and less anastomotic stricture formation. However, the most common postoperative complication is fecalith formation secondary to a rectal blind pouch or remnant colorectal septum [6]. Rarely, the fecalith could lead to the area of ischemic colorectal pressure ulcers, eventually causing localized ischemic necrosis and colorectal perforation [7,8]. Perirectal abscess and rectal fistula formation are observed after rectal perforation [9]. A wide range of treatment options is available, from a conservative treatment option to a wide intestinal resection and stoma in the treatment of rectosigmoid perforations. Innovative interventional endoscopic techniques have accelerated the development of endoscopic closure techniques such as clipping, stenting, suturing, gluing and Endoscopic Vacuum Therapy (EVT). These minimally invasive closure techniques have revolutionized the management of GI defects, providing a more affordable alternative to surgery with less morbidity and resource utilization. However, the real debate is what treatment will be applied to whom [10,11].

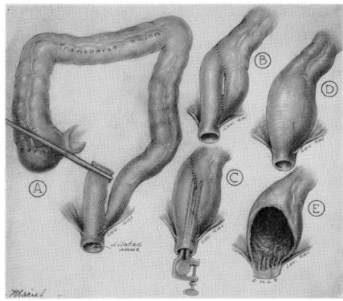


Figure 1: A. Following resection of the aganglionic portion of the colon, the proximal end is placed in the presacral space and the end of the colon anastomosed to the posterior wall of the rectum through an incision in the posterior wall of the rectum made transversely through the dilated anus. (B) Anastomosis from the proximal end of the rectum to the side of the adjacent colon is created, employing the open anastomosis technique. (C) The remaining colo-rectal septum is obliterated by means of a spur-crushing clamp which is inserted from below by an assistant while the surgeon is completing the proximal anastomosis. This permits placement of the clamp under direct vision before the anterior row of the anastomosis has been completed. The clamp is tightened in position and obliterates the remaining colo-rectal septum within a period of three to five days. (D) The appearance of a completed anastomosis. (E) The interior of the completed anastomosis.

Case Presentation

We present a case of a massive faecalith with refractory perirectal abscess and fistula occurring 50 years after Duhamel operation for Hirschsprung's disease. A 51-year-old man had undergone a Duhamel operation for Hirschsprung's disease at one year of age. In this case, he was admitted to our hospital because of intense abdominal pain in the lower left quadrant, constipation, tenesmus, and fever (38C). Preoperative CT examination revealed the abscess in the lesser pelvis between the blind rectal pouch and the sigmoid colon, which was accompanied by irregular rectal pouch wall thickening with a large faecalith, less pronounced sigmoid colon wall thickening with diverticulas and air leak in mesorectum (Figure 1). Presumably, a faecalith in the blind rectal pouch eroded into the rectum and developed a mesorectal abscess and pelvirectal fistula. During the colonoscopy fecalith was identified (Figure 3). However, removal of the fecalith was unsuccessful. The patient subsequently underwent manual disimpaction under intravenous general anaesthesia in the operating room. The colonoscopy revealed a 2cm diameter defect of the superior rectal pouch wall, complicated by the mesorectal abscess. EVT was used to treat defect in the rectum wall. The technique involves placement of the endosponge at the intra-luminal portion of the defect or more commonly deep into the surrounding contaminated cavity via an over-tube introduced through a standard endoscope. The endosponge was connected to a vacuum device with a constant pressure of 125-150 mmHg. In total, 3 sponges for EVT were changed during the period of 1 week to prevent the growth of the tissue into the sponge, avoid the loss of suction forces and allow inspection of the abscess cavity. After one week, the site of the rectal pouch defect was covered with granulation tissue showing an excellent healing (Figure 3). Patient was checked one month after discharge. Antibiotic therapy was completely stopped, and he had no fever, pain or bleeding during defecation. Control colonoscopy was done showing a possible fistula in the site of the defect. As a reason, pelvic MRI was performed two months after discharge to clarify diagnosis - no fistula or rectal pouch wall defects were shown, only irregular left lateral wall thickening, mild mesorectal fat and mesorectal fascia fibrosis were left (Figure 2).

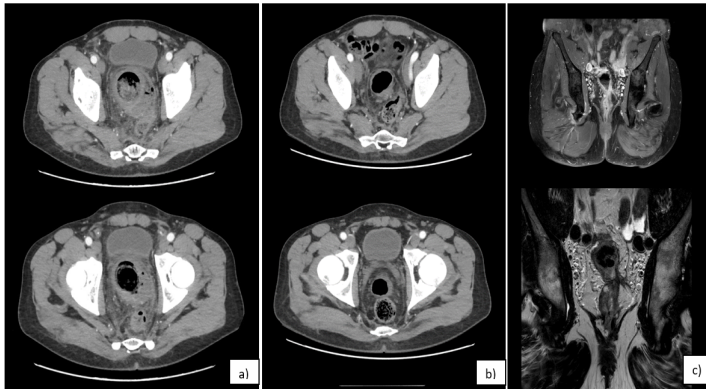


Figure 2: a) Abdomen CT scan before treatment: fecalith in the rectal pouch with perirectal abscess and air leak in mesorectum. b) Abdomen CT 2 weeks after treatment: no abscess or air leak in mesorectum c) Pelvic MRI scan 2 months after treatment: rectal pouch is fecalith free, no wall defect/fistula or mesorectal abscess suspected.

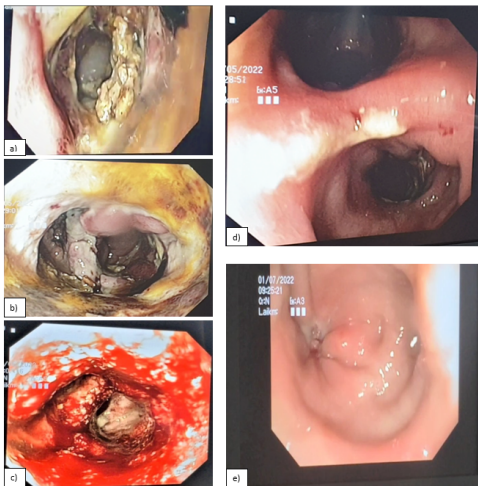


Figure 3: a) Pre-treatment endoscopy: a fecalith in rectal pouch b) Endoscopy after the removal of fecalith: abscess cavity c) 1 week after EVT treatment: rectal pouch defect with granulation tissue d) Reconstruction of the rectal pouch after Duhamel operation e) 1 month after EVT treatment: a possible fistula in the site of the defect.

Discussion

We presented a rare case of perirectal abscess and fistula secondary to fecalith eroding rectal pouch wall after Duhamel operation performed 51 years ago for Hirschsprung's disease. Constipation and faecal impactions as obstructive symptoms are common complications of Duhamel operation [12]. Obstructive symptoms following Duhamel reconstruction could be related to a transition zone pull-through or a large dysfunctional aganglionic

pouch. The aganglionic portion causes constipation, and as constipation builds, the pouch distends. This would explain why some patients have had multiple resections of the same pouch or septum, with recurrent symptoms [13]. Faecal impaction if left untreated, can lead to the hardening of stools and the formation of fecalith [14]. Fecaliths are generally considered a benign condition that can often be resolved with non-surgical interventions, including mineral oil enemas, manual disimpaction. Rarely, fecaliths can lead to more severe complications. A stercoral perforation of the rectum due to a faecaloma is a rare disease with a high mortality rate [15]. Persistent intestinal obstruction leading to bowel wall compromise or perforation is a surgical emergency, which usually requires operation [16]. To our knowledge, only one case of a massive fecalith with refractory perirectal abscess and recto-vesical fistula occurring 37 years after Duhamel operation was presented by Nakayana, et al. [17]. On this issue, resection of the common septum using endo-GIA stapler and removal of the faecalith were performed. The outcome of the surgical treatment was similar to our showing disease-free at one year follow-up. Innovations in endoscopic closure devices and techniques have provided a promising less invasive approach to managing GI perforations, leaks, and fistulas with good patient outcomes [11]. Self-Expandable Metal Stents (SEMS) or various endoscopic clips may be used in early esophageal fistulas and anastomotic leaks; however, these methods cannot be applied for the treatment of chronic fistulas and abscesses, especially after colorectal surgeries. Endoscopic drainage of abscess following perforation can be managed with EVT. The main advantage of this procedure is ensuring continuous drainage of the abscess cavity and acceleration of the formation of granulation tissue resulting in a reduction of the abscess cavity. EVT was invented and introduced into clinical practice approximately 20 years ago and its popularity in the treatment of post-operative anastomotic leakage has grown in recent years [18]. It has been shown to be effective in managing anastomotic leaks following esophageal as well as colorectal surgery [19,20]. In the review, Arezzo et al evaluated the long-term efficacy of endoscopic vacuum therapy for the treatment of anastomotic leaks after colorectal surgery [21]. EVT was applied in 14 patients with a success rate of 79%. The median time to complete healing was of 40.5 days. In our case, the duration of treatment was shorter. The size of the cavity was relatively small, and the patient did not have any comorbidities. Therefore, we believe healing process was quick in our case.

A spectrum of entities can mimic abdominopelvic abscesses and it may pose a diagnostic challenge [22]. Types of entities include neoplasms, diverticula, congenital variants, postoperative changes. In our case, the cause of abscess formation was firstly differentiated with complicated sigmoid diverticulitis and accompanying mesorectal abscess. There were several diverticula, though their wall and sigmoid wall thickening were less

pronounced compared to rectal pouch wall thickening. All typical complicated diverticulitis signs were found in our case: pericolic stranding, disproportionately prominent compared to amount of bowel wall thickening, segmental thickening of bowel wall, abscess (that contains fluid and gas) formation [23]. However, all these radiological signs were more pronounced in the rectal pouch suggesting it can be the primary inflammatory source, and sigmoid diverticulitis - secondary.

Conclusions

A rectal blind pouch remains a significant risk factor for late complications after Duhamel operation. We recommend strictly controlled defecation using enema once a month to avoid severe constipation. Otherwise, this may lead to fecalith formation and subsequent perforation of rectal pouch. Such a condition may pose diagnostic challenges in Radiology. The EVT treatment of perirectal abscess and fistula could be one of the most promising applications of vacuum-assisted closure in coloproctology. Treatment strategy should be adopted individually.

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