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ENDOSCOPIC ULTRASOUND-GUIDED DRAINAGE OF EXTRALUMINAL COLORECTAL MUCOCELE USING A LUMEN-APPOSING METAL STENT: A NOVEL APPROACH

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ABSTRACT

Colorectal mucoceles are rare mucinous cysts arising in the gastrointestinal tract and presenting diagnostic and therapeutic challenges. We report the first successful treatment of an extraluminal colorectal mucocele using a lumen-apposing metal stent (LAMS). A 65-year-old male with a known medical history of diverticulitis treated with a partial colectomy presented with constipation and low back pain. Computed tomography scan (CT) revealed a 5.4 cm cystic structure adjacent to the rectal anastomosis. Endoscopic ultrasound (EUS) found a subepithelial lesion with an "onion peel" appearance, and aspiration cytology confirmed a colo-colonic mucocele.

Management consisted of EUS-guided placement of an 8 × 8 mm LAMS, allowing for drainage, irrigation, and suctioning of the cavity. Complete resolution was achieved without recurrence over a 2-year follow-up period. This case demonstrates the potential of advanced endoscopic techniques, particularly LAMS, in managing extraluminal colorectal mucoceles. While further research is needed to compare the efficacy of LAMS with other treatment modalities, this procedure offers a minimally invasive alternative to surgery for certain patients.

KEYWORDS

Colorectal mucocele, endoscopic ultrasound, stent

LEARNING POINTS

- Novel approach to treat a rare case of colorectal mucocele using a lumen-apposing metal stent.

INTRODUCTION

Mucoceles are mucus-filled cysts typically seen in the oral cavity mainly on the inner aspect of the lower lip. In the gastrointestinal (GI) tract, they are rare, most commonly appearing in the appendix and much less often in the rectum[1]. Descriptions of colorectal mucoceles in the literature are scarce and only consist of case reports. Usually, these lesions are asymptomatic, but they can present with a variety of GI symptoms such as abdominal pain, tenesmus, constipation and, if large enough, with signs of obstruction[1,2]. We describe

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the first case of an extraluminal colorectal mucocele treated with a lumen-apposing metal stent (LAMS).

CASE DESCRIPTION

A 65-year-old man with hypertension, diabetes mellitus, and a history of diverticulitis treated with partial (sigmoid) colectomy presented to the emergency department with a 3-day history of lower back pain. The pain was sharp, intermittent, and associated with constipation, but the patient was passing flatus. He denied other GI symptoms. He was hemodynamically stable, afebrile, and had mild left lower quadrant tenderness on physical exam. Labs showed mild leucocytosis (12.16 $\times 10^3$/µl, normal: 4.8 to 10.8 $\times 10^3$/µl) and a lipase of 142 (normal: 7-60 U/l). A computed tomography (CT) scan of the abdomen and pelvis with intravenous contrast revealed a rectal anastomosis with an adjacent mural fluid collection or cystic structure measuring up to 5.4 cm *(Fig. 1)*. The patient underwent a diagnostic lower endoscopic ultrasound (EUS) which showed a colo-colonic anastomosis at 12-15 cm from the anal verge with a 5 cm subepithelial lesion bulging into the lumen, occupying 50-75% of it *(Fig. 2)*. The scope advanced proximally without resistance. Using a linear echoendoscope, the subepithelial lesion appeared anechoic with an onion peel appearance *(Fig. 2)*. Aspiration yielded 3 ml of thick mucoid clear liquid, and cytology confirmed a colo-colonic mucocele. After thorough consultation with the patient about available treatment options, the patient agreed to return for LAMS placement to ensure proper drainage of the cavity with further irrigation and suctioning of the collection cavity.

Two weeks later, the patient underwent a lower EUS with LAMS placement for drainage. The subepithelial lesion, measuring 6.6 × 5.3 cm, was located, and an 8 x 8 mm cautery-assisted LAMS was deployed under Doppler guidance. Clear, thick mucoid material drained through the LAMS lumen. The stent was dilated to 8 mm using a wire-guided controlled radial expansion balloon. A neonatal endoscope was used to access the collection cavity for irrigation and suction, achieving complete decompression and near resolution of the luminal narrowing *(Fig. 3)*. Two

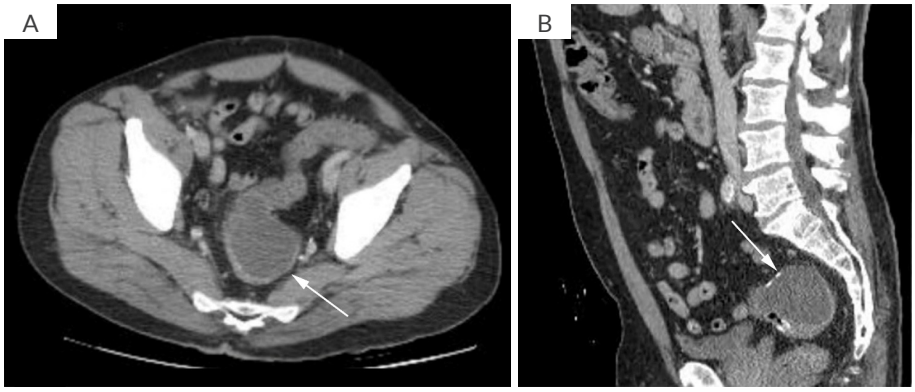


Figure 1. A computerized tomography scan of the abdomen and pelvis showing a rectal anastomosis with an adjacent mural fluid collection or cystic structure measuring up to 5.4 cm (white arrows). A) Axial and B) sagittal view.

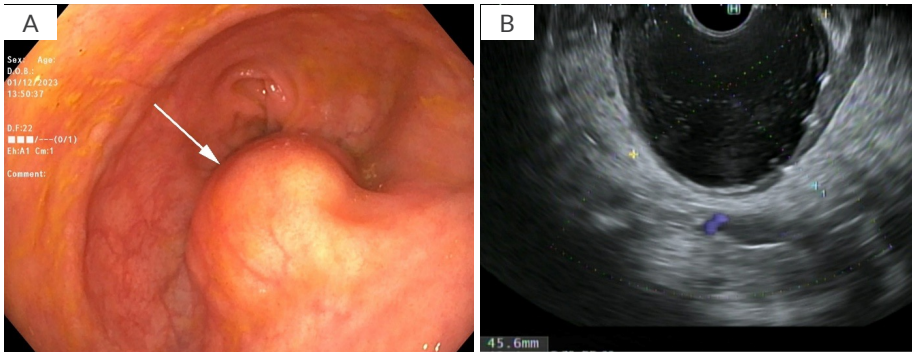


Figure 2. A) Subepithelial lesion bulging into the lumen at the colo-colonic anastomosis at 12-15 cm from the anal verge (white arrow). B) Endoscopic ultrasound showing a subepithelial anechoic lesion with an onion peel appearance.

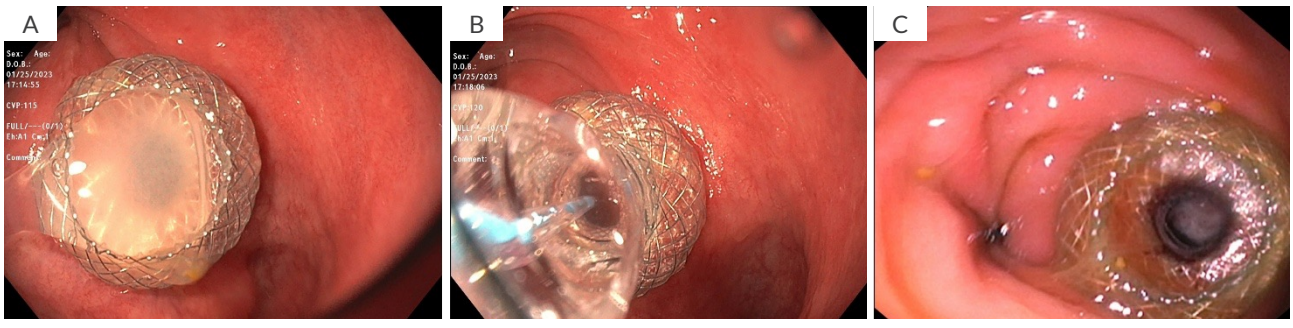


Figure 3. A) Colonoscopic image showing an 8 × 8 mm cautery-assisted LAMS draining thick mucus from the mucocele. B) LAMS dilation using a wire-guided CRE balloon. C) Mucocele cavity visualized through the dilated LAMS opening after irrigation and suction.
Abbreviations: LAMS, lumen apposing mucosal stent; CRE, controlled radial expansion.

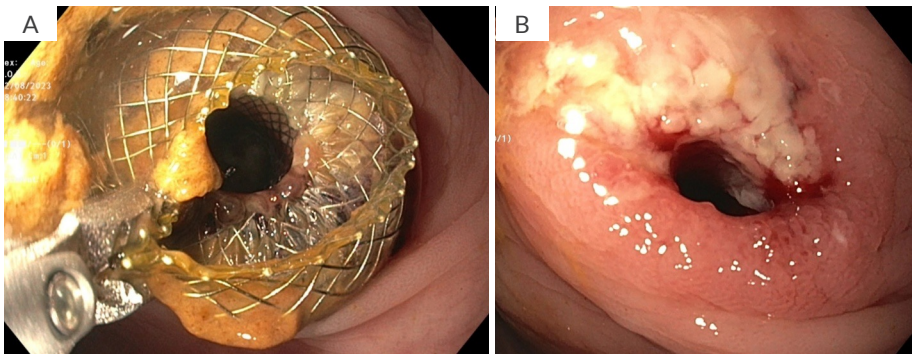


Figure 4. A) Removal of the lumen apposing mucosal stent (LAMS) using a forceps. B) Opening of the mucocele cavity after LAMS removal.

weeks post-procedure, sigmoidoscopy confirmed complete resolution of the mucocele, and the LAMS was safely removed *(Fig. 4)*. The patient was followed for 2 years without recurrence of symptoms or mucocele and no side effects.

DISCUSSION

As previously mentioned, colorectal mucoceles are extremely rare with both luminal and extraluminal lesions being described. Luminal colorectal mucoceles develop due to mucus build-up inside the bowel lumen and have been seen with closed loop obstruction either secondary to malignancy or inflammatory bowel disease[3]. Mucocele has also been reported after a Hartman procedure or a surgical end colostomy, after which, the mucus continuously secreted by the rectal mucosa, builds up in the distal stump due to an outlet obstruction like an anal stenosis[2,4,5]. Extraluminal mucoceles, similar to the lesion in our patient, have been attributed to either an anal gland obstruction, rectal mucosa invagination during surgical suturing or sequestration of a bowel wall segment during a colo-colonic anastomosis leading to the accumulation of mucinous secretions outside of the bowel lumen[1,3,6].

Patients can present with a variety of symptoms such as abdominal distention, rectal mass, constipation, pain, tenesmus, anal discharge or even obstruction if the mucocele is large enough to compress surrounding bowel portions. The most severe complication is rupture into the peritoneal cavity causing pseudomyxoma peritonei. A high index of suspicion is necessary to ensure diagnosis of a mucocele[2,3].

Furthermore, radiological studies can be helpful. A CT scan might show a dilated colon or rectum filled with hypodense fluid in the case of a luminal colorectal mucocele compared to a hypodense mass adjacent to the bowel, usually near a previous anastomosis site in an extraluminal mucocele[1,3-5]. Recently, EUS has been helpful in diagnosing extraluminal mucoceles which appear as homogenous, hypoechoic lesions with internal layering similar to an onion skin appearance[1,7]. The "onion skin" sign has been observed on transabdominal ultrasound for appendiceal mucoceles and has a sensitivity of 63% and specificity of 100%[7]. Further studies are needed to confirm if this applies as well to EUS in colorectal mucoceles.

Variable treatment modalities for colorectal mucoceles have been described, ranging from manual digital evacuation, CT guided aspiration and surgical resection[1-3]. EUS-guided drainage has been successfully used as a treatment modality, avoiding the need for surgical intervention[7,8]. There is currently very limited data concerning the recurrence rate of mucoceles or the effectiveness of different treatment options. Additionally, there are no established guidelines regarding which treatment modality to use based on the mucocele's location, aetiology or size. However, a few case reports have reported recurrence when aspiration was used alone[1]. Another study suggested that when the mucocele was larger than 2 cm, surgery should be considered due to premalignant potential in appendiceal mucoceles, however no such data exists in colonic mucoceles[9].

Based on our search results, there is no specific documented case of a colorectal mucocele treated with a LAMS. The LAMS is currently approved by the Food and Drug Administration for the drainage of pancreatic fluid collections in the presence of walled-off necrosis and can be used (off-label) for gallbladder, biliary and pancreaticobiliary drainage, improving access for surgically altered anatomy, luminal stricture treatment, gastroenteric fistulae creation, and intra-abdominal and pelvic abscesses as well as post-surgical fluid collection drainage[10]. In one case report EUS-guided transrectal drainage with balloon dilation and placement of plastic stents was used for the treatment of a rectal mucocele with successful results[8]. In our case, after discussing with the patient regarding the various treatment options and taking into account the size of the mucocele and the risk of recurrence when drainage is used alone, we decided to employ a LAMS to treat the mucocele. The LAMS allowed for efficient drainage, coupled with thorough irrigation and suctioning of the collection cavity reducing the risk of recurrence.

To our knowledge, this is the first reported use of EUS in diagnosing an extraluminal colorectal mucocele, combined with successful drainage using LAMS. This novel approach demonstrates the potential of advanced endoscopic techniques in managing colorectal mucoceles however further studies would be required to better explore the efficacy and outcomes of using LAMS for this condition compared to other treatment modalities.

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