Multi-Colitis Cystica Profunda: A Case Report

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Colitis cystica profunda (CCP) is a benign disease characterized by mucin-filled cysts beneath the muscularis mucosa.[1] CCP can present in a localized form with a polypoid lesion, or as a more diffuse process involving a variable length of the rectal mucosa or colon.[2] Cysts, which may be quite large, are localized to the rectosigmoid region and are usually 6 to 7 cm from the anal verge. CCP has been observed in patients between 30 and 40 years of age.

Signs of CCP include rectal bleeding, mucorrhea, diarrhea, and colonic obstruction, similar to other associated disorders.[3] CCP is associated with a variety of ulcerating diseases including inflammatory bowel disease (ulcerative colitis [UC] and Crohn's disease), infectious colitis, rectal prolapse, solitary rectal ulcer, and diverticulitis, among others.[4-6] CCP can resemble adenoma, adenocarcinoma, lipoma, endometriosis, neurofibroma, pseudopolyps, and pneumatosis coli.[7] However, the etiology of CCP is not yet well-defined.

Endoscopy and barium studies can reveal CCP lesions. Endoscopic ultrasound (EUS) can be used to identify cysts in the rectal wall. Computerized tomography (CT) scan or magnetic resonance imaging can show noninfiltrating submucosal masses, loss of perirectal fatty tissue, and thickening of levator ani muscles.[2] Diagnosis depends on histologic examination.

A 29-year-old woman was admitted to our hospital for left lower quadrant abdominal pain with abdominal distension that had occurred for 1 year. She had no fever, chills, rectal bleeding, or nausea, but had occasional vomiting. She had no family history of any colorectal disease. Laboratory tests for complete blood count, blood chemistry, liver function, stool, and tumor markers were normal. Colonoscopy revealed two submucosal tumors, 15 mm and 25 mm in diameter, which located about 12 cm and 7 cm from the anal verge, respectively [Figure 1a]. CT scan displayed that the rectum was segmental thick and convex soft tissues in the cavity with internal nodular calcification [Figure 2a]. EUS showed a submucosal mixed cystic-solid echo with hypoechoic nodules that had thick mucosal layers [Figure 2b].

Pathologic analysis of a deeply biopsy specimen and EUS guided-fine needle aspiration (EUS-FNA) was inconclusive. Instead, we performed endoscopic submucosal dissection (ESD) of one lesion to obtain a complete tissue sample. Followed the EUS, we performed one masses by ESD. In the process of resecting the larger (15-mm) mass, a yellow-white viscous substance was released. Cytology of cast-off cells from the rectum revealed normal subleaf nuclear granulocytes,

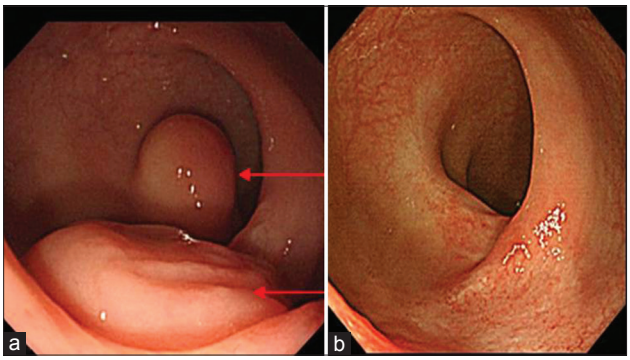

Figure 1: (a) Colonoscopy revealed two submucosal tumors (indicated by red arrows), (b) smooth, normal-appearing rectal mucosa 1 year after endoscopic submucosal dissection.

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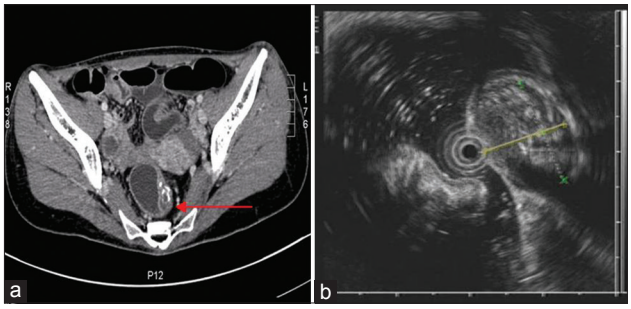

Figure 2: (a) Computerized tomography noted the presence of segmental thickening of the rectum with convex soft tissue density (red arrow), (b) endoscopic ultrasound revealed a submucosal mixed cystic-solid echo with thick mucosal layer.

lymphocytes, histiocytes, columnar epithelial cells, and squamous epithelial cells. Pathological analysis showed cystica profunda. Gross pathology of the masses removed by ESD revealed surrounding mucus and fibrosis, with partial calcification [Figure 3]. No signs of malignancy were evident. Moreover, we removed another mass by ESD 6 months later. A colonoscopy performed 1 year after the ESD operation demonstrated that the patient's rectal mucosa was normal and smooth [Figure 1b].

So far, this is a rarely reported case of CCP lesions within the rectum, because the pathologic analysis was very difficult. The patient with multiple CCP was seldom reported. Recently, a case of a single polypoid CCP lesion was reported in association with adenocarcinoma.[7] The sample of endoscopic biopsy or FNA is not enough. In this case, ESD is a technology that can remove the whole mass that can help pathologist analysis.

Some evidence suggested that CCPs was connected with UC. However, this patient had no history of UC. Hence, we should advise high fiber diet and avoid straining while defecating. Biofeedback therapy can be helpful and pharmacological therapies include lubricants, bulking laxatives, sucralfate, and hydrocortisone enemas.[7] However, patients with rectal prolapse should be considered for surgical treatment by resection and suture rectopexy.[8]

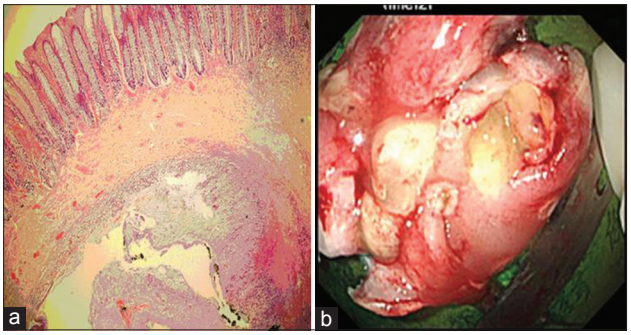

Figure 3: (a) Histological examination of biopsied specimens confirmed submucosal mucous cysts. (H and E, original magnification ×40), (b) Gross pathology of the masses removed by endoscopic submucosal dissection revealed surrounding mucus and fibrosis, with partial calcification.

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Conflicts of interest

There are no conflicts of interest.

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