

Proctitis Cystica Profunda as Submucosal Tumor in the Rectum

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Abstract

Proctitis cystica profunda is mucous-filled cystic tumor with benign epithelial lining. It has known as a rare non-neoplastic condition in the submucosal layer, especially in the rectum. Herein, we report a cases of proctitis cystica profunda as submucosal tumor in the rectum. Fifty-four-year-old female, who had a submucosal tumor at the rectum, underwent transanal excision. It was 2.0x2.0 cm sized cystic tumor, filled with dark brown colored, mud-like material. Diagnosis was proctitis cystica profunda. She discharged at the 4th postoperative day without postoperative complication.

Keywords: *Benign tumor, Rectum*

INTRODUCTION

Proctitis cystica profunda is a rare, benign, non-neoplastic condition that usually recognized as a mass in the rectum, often confused with rectal carcinoma (1, 2). It has known, closely associated with solitary rectal ulcer syndrome and rectal prolapse. However, its exact etiology remains unclear. It is diagnosed histologically by the presence of mucous-filled cysts with benign epithelial lining in the submucosal layer (3). Although it is uncommon condition, proctitis cystica profunda has to be considered to differentiate form other common rectal neoplastic conditions. Herein, we report a cases of proctitis cystica profunda as submucosal tumor in the rectum.

CASE REPORT

Fifty-four-year-old female was admitted with a submucosal tumor at the rectum. One year ago, she visited emergency room with abruptly onset, abdominal pain on periumbilical region. Vital signs were stable. Blood pressure was 120/70 mmHg. Pulse rate was 55 per minute. Respiration rate was 20 per minute. Body temperature was 36.5 degree Celsius. She had a history of hypertension. She had no history of constipation and rectal prolapse. Symptom was improved with conservative management. She discharged emergency room at that day.

Five-days later, she revisited outpatient clinic for follow-up for previous abdominal pain. Also, she complained a palpable mass on the epigastrium. Further evaluation studies were undergone. Incidentally, a submucosal rectal tumor was identified. On the digital rectal examination, the cystic tumor was soft, round palpable submucosal tumor at 3-4cm from the anal verge, 5 o'clock direction in the rectum. Computed tomography showed a homogenous, non-enhanced cystic tumor, with 4cm diameter, in front of the coccyx, in the rectum (Fig 1). There was no remarkable finding at the abdominal wall, peritoneum and the intra-abdominal organ. Colonofiberscopy showed a 4x4cm sized round elevated lesion covered with normal mucosa at the 5cm from the anal verge (Fig 2). Cushion sign was positive. Endoscopic ultrasound showed a round, well-demarcated homogenous hypoechoic mass arising from the muscle propria layer (Fig 3). The gastroscopy showed diffuse atrophic gastritis and gastric erosion at the greater curvature side of the low body. She underwent transanal excision. The specimen was 2.0x2.0 cm sized cystic tumor, filled with dark brown colored, mud-like material (Fig 4). Diagnosis was proctitis cystical profunda. She discharged at the 4th postoperative day without postoperative complication.

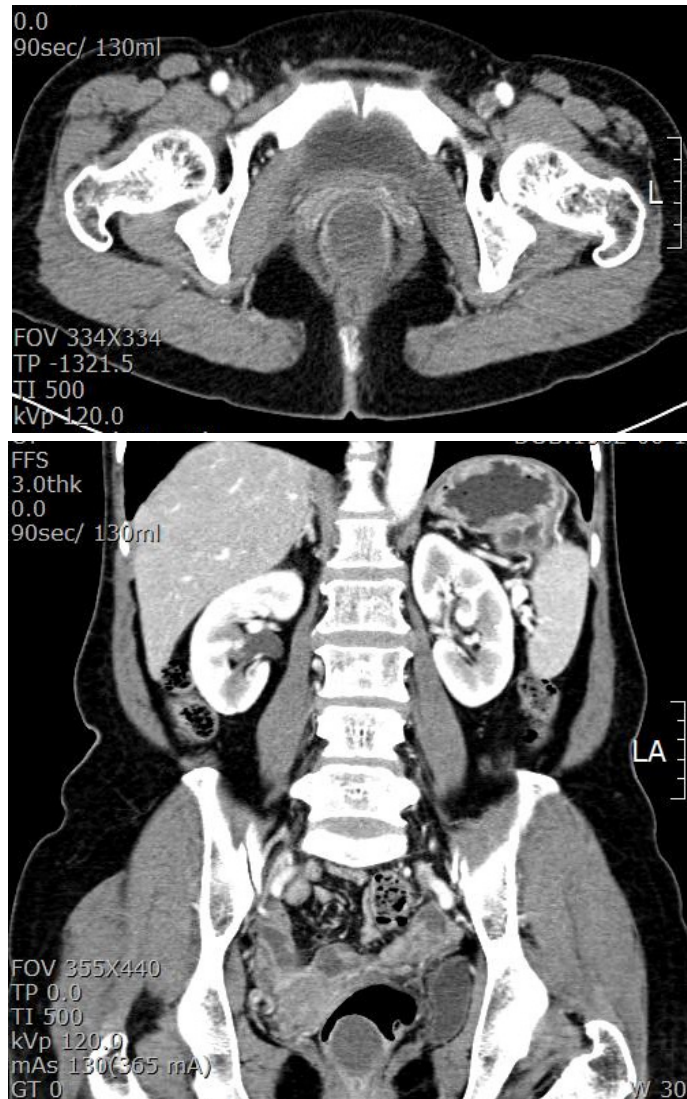


Fig 1. Computed tomography showed a homogenous, non-enhanced cystic tumor, with 4cm diameter, in front of the coccyx, in the rectum (A,B).

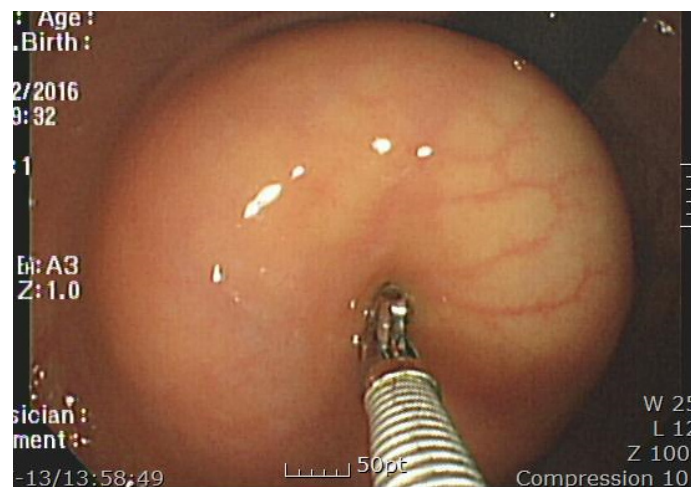


Fig 2. Colonofiberscopy showed a 4x4cm sized round elevated lesion covered with normal mucosa at the 5cm from the anal verge. Cushion sign was positive.

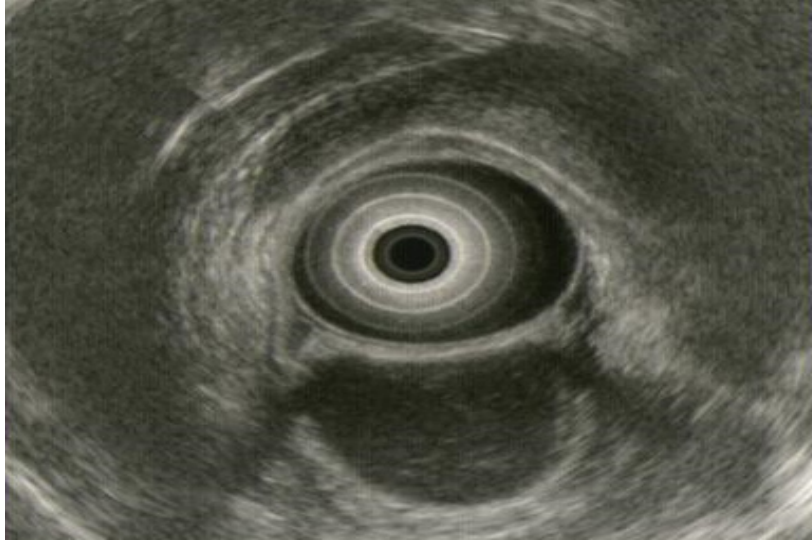


Fig 3. *Transrectal ultrasonography showed a round, well-demarcated homogenous hypoechoic mass arising from the muscle propria layer.*

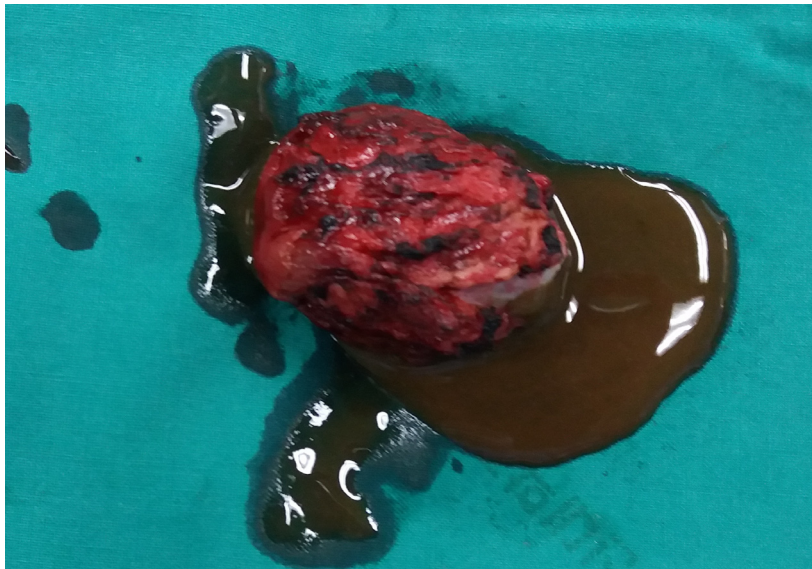


Fig 4. *Specimen showed was 2.0x2.0 cm sized cystic tumor, filled with dark brown colored, mud-like material.*

DISCUSSION

There are many variable conditions diagnosed as a submucosal tumor in the rectum, such as lipoma, leiomyoma, hemangioma, gastrointestinal stromal tumor, carcinoid tumor and carcinoma (4). Also, proctitis cystica profunda is usually recognized as a submucosal tumor, especially in the sigmoid colon and the rectum. Although it is uncommon, proctitis cystica profunda has to be considered as possible condition for a submucosal tumor in the rectum.

Although colonoscopy provides excellent visualization, enables endoscopic biopsy, it has limitations to evaluate for a submucosal tumor in the rectum, exactly.

In contrast, imaging studies, such as transrectal ultrasonography (TRUS), computed tomography (CT), magnetic resonance imaging (MRI), allows evaluating perirectal tissues and pelvic organs in addition to the full-thickness of the rectum. In particular, MRI has known as the best investigative modality to evaluate a submucosal tumor, can help achieve accurate preoperative diagnosis and facilitate the appropriate management (4). In this case, CT showed the submucosal tumor which was cystic and relatively large with 4cm diameter, in front of the coccyx. A presumptive diagnosis was a tailgut cyst.

Although the pathogenesis of this condition is not clearly established, proctitis cystica profunda is

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considered to associate with a defecation disorder, a solitary rectal ulcer syndrome consequent to rectal prolapse. Excessive straining at defecation, ischemia and ulceration of the prolapsed rectal mucosa is thought to be possible etiologic causes (3,5,6). During healing of the ulcer, the glandular epithelium extended into the submucosa. And Necrosis of the submucoal lymphoid aggregations follows (1,2). In this case, the patient has no history of a defecation disorder, a rectal prolapse or a rectal trauma.

Symptoms include blood and mucus in the stool in over half of reported cases. Diarrhea, tenesmus, abdominal pain, or rectal pain are also reported (1, 2). The common location of the lesion is the rectum, ranged from the dentate line to 12cm (mean 5.5cm). The size of the lesion varied from 2 to 12cm (mean 3cm). It usually located at the anterior wall of the rectum (2). In this case, the patient visited the emergency room abruptly onset-periumbilical abdominal pain. But, the submucosal tumor was identified incidentally during further evaluation study. Correlation between abdominal pain and the submucosal tumor, proctitis cystica profunda, was controversial. The submucosal tumor was located at the rectum from the anal verge 3-4cm distance, with 2cm sized-diameter.

Histologically, it is characterized with features of early changes of replacement of the normal lamina propria by fibroblasts arranged at right angles to the muscularis mucosa. The muscularis mucosa is thickened. There is a reactive hyperplasia in the overlying epithelium with goblet-cell depletion. Mismatched epithelial elements and lamina propria may be seen in the submucosa and their cystic dilatation. The cystic lesion is filled with mucus (2).

Once the diagnosis of proctitis cystica profunda has been reached or should it be suspected, surgical treatment with local excision can be attempted. However, a 25 percent rate of recurrence has been reported (7). If it is associated with rectal prolapse, corrective surgical approach for rectal prolapse has to be considered at same time of local excision.

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