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CASE REPORT

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Solitary rectal ulcer syndrome in a young adult – A surgeon's dilemma with rectal carcinoma

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Abstract

The presentation of solitary rectal ulcer syndrome is very similar to a wide variety of conditions including inflammatory bowel diseases, ischemic colitis and rectal carcinoma. Histopathological examination comes as an important tool for its diagnosis. Hence, high index of suspicion is required for early diagnosis of this rare condition.

KEYWORDS

bleeding per rectum, constipation, rectal carcinoma, solitary rectal ulcer

1 | INTRODUCTION

Solitary rectal ulcer syndrome (SRUS) is a rare, benign and chronic disorder characterized by a combination of symptoms, endoscopic findings and histological abnormalities.[1] It was first described by Cruveihier[2] in 1829, but it became widely recognized only after review of clinical and pathological features of this condition by Madigan and Morson in 1969.[3] SRUS is a misnomer because the endoscopic findings are not just limited to a single rectal ulcer as suggested by the name. In fact, numerous ulcers are found in 40% of the patients while solitary ulcer, as mentioned in the name, is found in only 20% of the patients. Rest of the patients have lesions ranging from mucosal erythema alone to single or multiple ulcers and broad-based polypoid lesions.[4]

The incidence of SRUS has been estimated to be 1 in 100,000 individuals per year, affecting both males and females with slight female predominance.[5] The median age at diagnosis is 48 years with a range of 14–76 years.[4] Clinical features includes rectal bleeding, copious mucus discharge, prolonged excessive straining, perineal and abdominal pain, feeling of incomplete defecation, constipation and rarely, rectal prolapse[6] with the most common symptoms being rectal bleeding (56%), straining with defecation (28%) and pelvic fullness (23%).[7] However, as much as 26% of the patients can be asymptomatic.[4,7–9]

Here, we present a case of SRUS in a young adult mimicking as rectal carcinoma.

2 | CASE PRESENTATION

A 34-year-old male patient presented to surgical out patient department with a history of recurrent per rectal bleeding which has increased in intensity for past six months. The bleeding was painless, scanty, fresh, usually mixed with stool and at the end of defecation. Bleeding was not associated with tenesmus, fever, abdominal pain or self-digitation. He had increasing frequency of constipation for past one year, for which he has been taking oral laxatives. He also had unintentional weight loss of five kgs in the past six months. There was no history of diabetes mellitus, hypertension or previous allergic reactions to

any drugs or foods. There was no history of surgical intervention in the past. There was no history of gastrointestinal malignancies among his family members.

On his general examination, vital signs were within the normal limits. On his abdominal evaluation, the abdomen was soft, without tenderness and with normal bowel sounds. Proctoscopy examination revealed an irregular broad-based ulcerated mass in the anterior wall of the rectum. Fecal occult blood test was negative. His laboratory findings were within the normal range (hemoglobin level 14.6 g/dl, white blood cell count 7900/mm^3 and platelet count 171,000/mm^3). His liver function tests, erythrocyte sedimentation rate, C-reactive protein and coagulation profile were also normal.

He was advised for colonoscopy which showed a hemorrhagic and circumferential ulcerated mass with edema in the anterior rectal wall located five cms from the anal verge. The ulcerated mass was taken for biopsy. He also underwent abdominopelvic contrast enhanced computed tomography (CECT) scan which exhibited an area of rectal wall thickening with perirectal fatty infiltration and enlargement of multiple small perirectal lymph nodes. For further evaluation, he underwent magnetic resonance imaging (MRI) pelvis which showed a similar finding of few subcentimetric perirectal lymph nodes suggestive of reactive lymph nodes. On further evaluation of tumor markers, carcinoembryonic antigen (CEA) level was 2.71 ng/ml (normal: 0–5.0 ng/ml), carbohydrate antigen 19.9 (CA19.9) −0.8 IU/ml (normal <37.0 IU/ml), antinuclear antibodies (ANA) (CLIA method)-7.82 AU/ml (normal: 32 AU/ml). His biopsy revealed a central ulcerated lesion accompanied by surrounding edema and inflammatory infiltrates, not invading the muscle layer, suggestive of SRUS. Along with mucosal ulceration, there was crypts distortion and hypertrophy of muscularis mucosa (Figure 1). There was no evidence of dysplasia and malignancy. Three months after conservative management with stool softener and pain control, clinical symptoms of patient improved. Follow-up colonoscopy showed that the lesion markedly improved with remnant ulcerative scarring.

3 | DISCUSSION

The average time duration between appearance of symptoms and correct diagnosis is 5 years in SRUS, ranging from 3 months to 30 years in adults, which, fortunately in pediatric patients is slightly less (1.2–5.5 years).[10] Patients usually present with fresh (bright) rectal bleeding, copious mucous discharge, tenesmus, perineal and abdominal pain and sometimes, with rectal prolapse as well.[6] These signs and symptoms are consistent with the history of prolonged excessive straining, constipation or abnormal defecation, including self-digitation to evacuate the impacted stool or to reduce the rectal prolapse. Although the underlying etiology and pathophysiology of SRUS is not well understood, it has been hypothesized that chronic damage to the rectal mucosa from direct trauma or local ischemia as a result of long-standing tenesmus, straining during constipation, intussusception of the rectal mucosa and inflammation from hard stool or digitation maneuvers may play a role.[11] It has been suggested that paradoxical contraction of puborectalis muscle during defecation in people with pelvic floor dyssynergia also leads to the final common pathogenic mechanism of overt or occult rectal prolapse over time, causing ischemic changes and ulceration in the rectal mucosa. This is supported by the anorectal physiology studies which show that 25%–82% patients with SRUS may have dyssynergia in them.[12] Among these different facets of multifactorial causation of SRUS, excessive straining during defecation and mucosal inflammation due to hard impacted stool may only be the causes for development of SRUS in this patient.

Diagnosis of SRUS is based on clinical findings, proctosigmoidoscopy and histopathological examination, imaging investigations including dynamic magnetic resonance imaging and anorectal functional studies including manometry.[13] As already mentioned above, the name of this entity- "SRUS" is a misnomer and its presentation can also easily misguide a clinician to other common diagnoses that fit better in the clinical picture like Inflammatory Bowel Disease (Crohn's Disease and

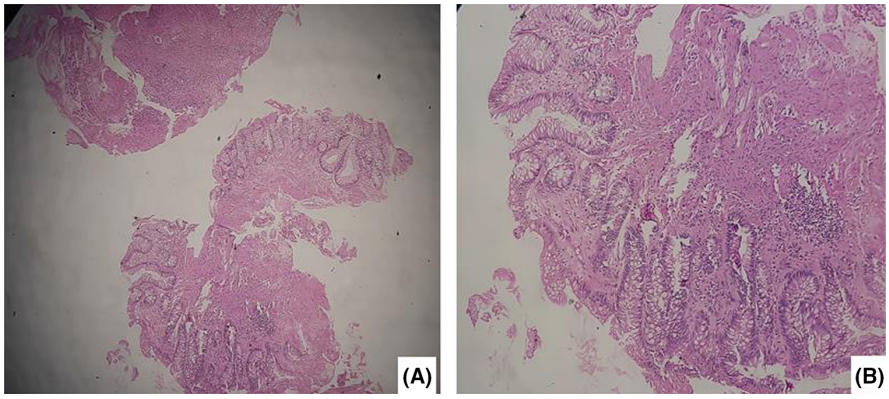

FIGURE 1 (A) (4× magnification): shows two tissue bits lined by colonic mucosal epithelium with mucosal ulceration, crypts distortion and hypertrophy of muscularis mucosae; (B) (10× magnification): shows mucosal ulceration with splaying of muscularis mucosae reaching up to the mucosal glands. Ectatic blood vessels are also seen along with inflammatory infiltrates

Ulcerative Colitis), ischemic colitis and rectal carcinoma. In a retrospective study of 80 patients with biopsy-proven SRUS, the variable macroscopic lesions included polypoid lesions in 44% (predominant lesion type in asymptomatic cases), ulcerated lesions in 29% and edematous, hyperemic, non-ulcerated mucosa in 27% cases.[4] Even the confirmatory histopathological examination sometimes fails to solve the issue of delayed diagnosis or misdiagnosis because of inadequate rectal biopsy and failure to recognize the histopathological features of the disease.[13] The ulcer characteristics of our patient that it was present 5 cm above the anal margin, on anterior rectal wall, is coherent with what has been reported in the literature (ulcers 0.5–4 cm in diameter, 3–10 cm above the anal margin).[3,7] Key histological changes present in SRUS include fibromuscular obliteration of lamina propria, hypertrophied muscularis mucosa with extension of muscle fibers upward (between the crypts) and glandular crypts abnormalities.[14] Other minor microscopic findings such as surface erosions, mild inflammation, distorted crypts and reactive epithelial atypia may lead to erroneous diagnoses such as IBD and cancer. But the differentiating histological markers would be diffuse collagen deposition in the lamina propria and abnormal smooth muscle fibers extensions.[15,16] Though the results of imaging investigations (CT and MRI) in our case directed the initial suspicion towards malignancy, it was however corrected by the biopsy findings, which helped in relatively early diagnosis of this condition.

Regarding treatment options, there are four basic pillars of treatment: conservative management (involving hygiene, dietary and behavioral modification), medical therapy, biofeedback therapy and surgery. Dietary and behavioral modifications are solely effective in patients with mild to moderate symptoms and without significant mucosal prolapse. This approach may not work in patients having advanced grade of rectal intussusception, extensive inflammation and fibrosis and/or reducible external prolapse. Moreover, conservative management can incorporate the use of drugs such as sucralfate, salicylates, corticosteroids, sulfasalazine, mesalazine and topical fibrin sealant for alleviation of symptoms.[17] For patients who are refractory to conservative measures, biofeedback appears promising, which corrects abnormal pelvic-floor behavior to reduce excessive straining with defecation. As a result, anorectal functions are improved in patients who exhibited dyssynergic defecation.[18] Surgery becomes a reliable option for patients who do not respond to above approaches (conservative and biofeedback) or have rectal prolapse. Surgical intervention includes ulcer excision and treatment of internal or overt rectal prolapse by performing mucosal resection (Delorme's procedure) or perineal proctectomy (Altemeier's procedure).[17,19]

In our case, the patient did not have rectal prolapse and was kept on conservative management with dietary modifications, appropriate lifestyle changes and drugs to soften the stool. Improvement in his clinical condition after three months emphasizes the necessity of early diagnosis of this condition in order to prevent the patient from unwanted medical and surgical procedures which always carry significant risks with them. For this, high degree of suspicion of SRUS as a differential diagnosis in above clinical picture is required from both surgeon's and pathologist's point of view.

4 | CONCLUSION

Conservative management with dietary modifications and lifestyle changes is one of the effective treatment modalities of SRUS. However, its presentation is very similar to a wide variety of conditions including IBDs, ischemic colitis and rectal carcinoma. Hence, high index of suspicion is required for early histopathological diagnosis of this condition.

AUTHOR CONTRIBUTIONS

Sunil Basukala (SB) and Ujwal Bhusal (UB) involved in conceptualization and supervision. Ujwal Bhusal (UB), Ayush Tamang (AT), Subodh Dhakal (SD), Shriya Sharma (SS), and Anuj Karki (AK) involved in writing—original draft. UB, SB, AT, SD, SS, and AK involved in writing—review and editing. All the authors read and approved the final manuscript.

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CONFLICT OF INTEREST

No conflict of interests.

DATA AVAILABILITY STATEMENT

All the findings are present within the manuscript.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editor-in-chief of this journal on request.

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