Solitary rectal ulcer syndrome presenting with rectal prolapse, severe mucorrhea and eroded polypoid hyperplasia: Case report and review of the literature

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DV Gopal, C Young, RM Katon. Solitary rectal ulcer syndrome presenting with rectal prolapse, severe mucorrhea and eroded polypoid hyperplasia: Case report and review of the literature. Can J Gastroenterol 2001;15(7):479-483. A case of solitary rectal ulcer syndrome in a 36-year-old woman presenting with severe, persistent mucorrhea and eroded polypoid hyperplasia as the predominant clinical features, who was ultimately noted to have symptoms of rectal prolapse, is presented. Endoscopically, she had multiple (50 to 60) small, whitish polypoid lesions in the rectum that were initially misinterpreted as being a carpeted villous adenoma, juvenile polyposis or atypical proctitis. The lesions were treated with argon plasma coagulation with resolution, but a solitary rectal ulcer developed. The patient then admitted to a history of massive rectal prolapse over the preceding six months and underwent surgical treatment. Severe mucorrhea as the presenting feature and the presence of multiple polypoid lesions consistent with a histological diagnosis of eroded polypoid hyperplasia make the present case unique.

Key Words: *Mucorrhea; Polypoid hyperplasia; Solitary rectal ulcer syndrome*

Syndrome de l'ulcère solitaire du rectum accompagnant un prolapsus rectal, une mucorrhée grave et une hyperplasie polypoïde érodée : exposé de cas et examen de la documentation

Voici le cas d'une femme de 36 ans souffrant du syndrome de l'ulcère solitaire du rectum, dont les principales manifestations cliniques se composaient de mucorrhée persistante grave et d'une hyperplasie polypoïde érodée; on a finalement noté des symptômes de prolapsus rectal. L'endoscopie a révélé la présence de multiples (50 à 60) petites lésions polypoïdes blanchâtres dans l'ampoule rectale; ces lésions avaient été faussement diagnostiquées comme un tapis d'adénomes villeux, une polypose juvénile ou une rectite atypique. Le traitement des lésions à l'aide de la coagulation au plasma-argon a porté fruit mais a été suivi de l'apparition d'un ulcère solitaire du rectum. La patiente a alors avoué qu'elle avait eu des antécédents de prolapsus rectal massif au cours des six mois précédents et qu'elle avait subi une intervention chirurgicale. La présence de mucorrhée grave comme premier signe de la maladie et de lésions polypoïdes multiples compatibles avec un diagnostic histologique d'hyperplasie polypoïde érodée donne au cas son caractère singulier.

Solitary rectal ulcer syndrome (SRUS) is a benign condition affecting the rectum that generally pursues a chronic course of rectal bleeding, mucorrhea and tenesmus (1). Severe mucorrhea as the major presenting symptom is uncommon and can lead to misdiagnosis. Endoscopically, only 35% of these rectal lesions are solitary ulcers, 22% are multiple ulcers and 44% show polypoid lesions with patchy, granular and hyperemic mucosa (2). A case of solitary rectal ulcer syndrome in a woman who presented with extreme mucorrhea and minimal rectal bleeding is presented. The

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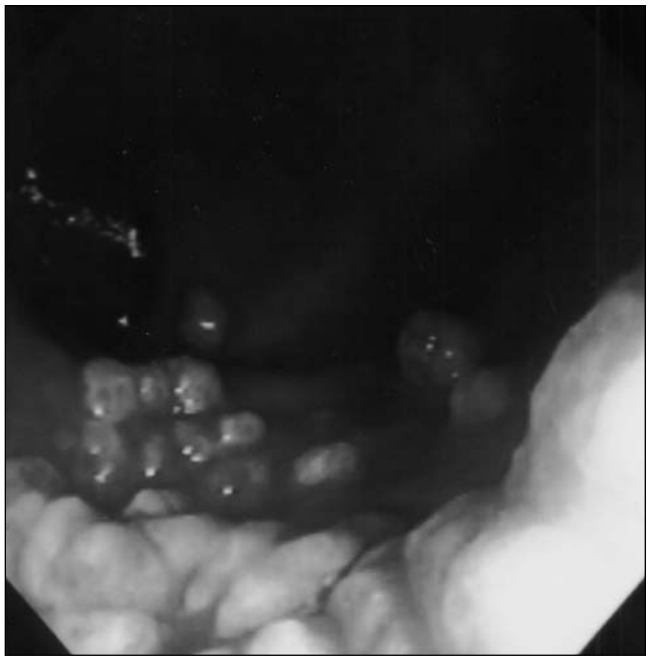

Figure 1) *Flexible sigmoidoscopy revealing multiple rectal polypoid lesions at 5 to 15 cm above the anal verge*

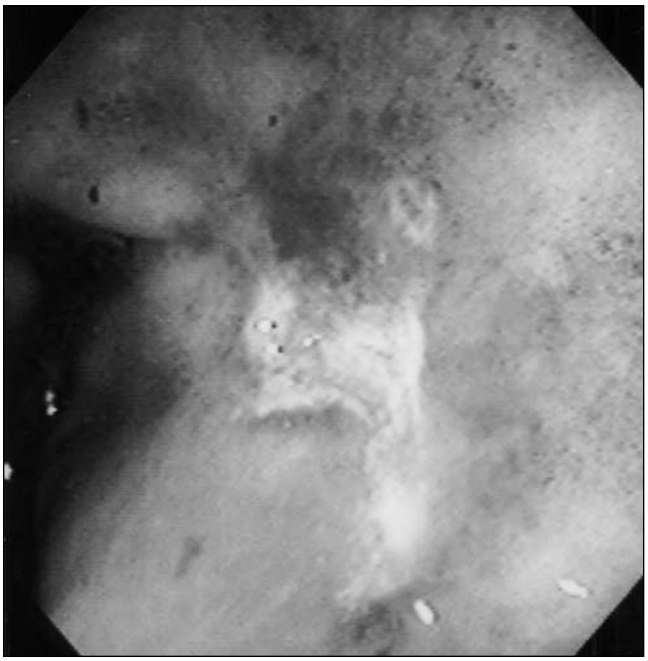

Figure 3) *Endoscopic findings of a solitary rectal ulcer after two treatments with argon plasma coagulation*

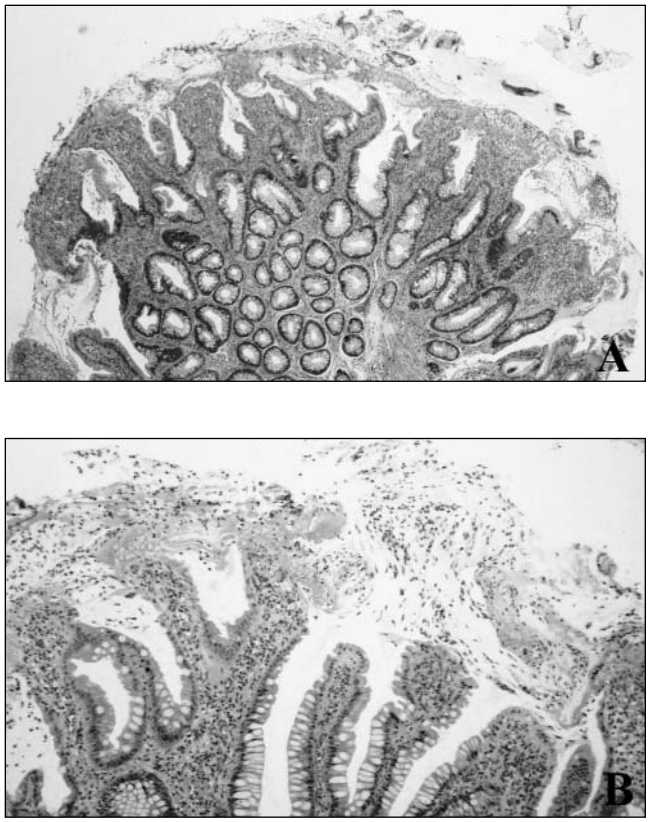

Figure 2) A *Histopathology of a polypoid lesion with surface erosion, reactive glands and fibrinomucinous exudate (hemotoxylin and eosin stain, original magnification ×40).* **B** *Close up of polypoid surface showing epithelial erosions and fibrinomucinous exudate (hemotoxylin and eosin stain, original magnification ×100)*

initial sigmoidoscopic appearance was compatible with eroded polypoid hyperplasia, which later progressed to solitary rectal ulcer. Misinterpretation of the significance of the nodular lesions led to the preliminary diagnoses of villous adenoma, atypical proctitis and juvenile polyposis, which caused a delay in the final diagnosis and appropriate therapy.

CASE PRESENTATION

A 36-year-old woman was referred to the authors' institution for endoscopic evaluation and management of abnormal findings noted on colonoscopy. She had developed symptoms of severe, persistent mucorrhea, left lower quadrant and perianal pain, and minimal rectal bleeding over a three-month period. The mucorrhea occurred day and night, necessitating the wearing of diapers, and forced the patient to quit her employment. Past medical history was negative for inflammatory bowel disease or colorectal malignancy. Physical examination was essentially unremarkable. Rectal examination revealed mucous discharge with minimal blood but no other significant findings. Initially, no obvious rectal prolapse was observed.

Initial colonoscopy at the referring hospital revealed numerous polypoid lesions in the rectum. The gross appearance suggested a carpeted villous adenoma, but biopsies disclosed nonspecific inflammation. Rectal installation of 5-aminosalicylate (5-ASA) (Rowasa; Solvay Pharmaceuticals Inc, USA) enemas failed to alleviate symptoms and the patient was referred to the authors' institution for atypical proctitis. Repeat flexible sigmoidoscopy done at the authors' institution showed 50 to 60 small, whitish polypoid lesions 5 to 15 cm from the anal verge in

the rectum only (Figure 1). Biopsies taken of six representative polyps showed inflammation of the lamina propria with associated surface erosions consistent with inflammatory, juvenile-type polyps versus hamartomatous polyps (Figures 2A,2B). On two follow-up occasions, these suspected juvenile polyps were treated with argon plasma coagulation (APC) (ERBE APC 300; ERBE USA Inc, USA) treatment (3); however, the mucorrhea persisted. On her third follow-up flexible sigmoidoscopy, the nodular lesions were no longer present, but she had a 1×1.5 cm solitary rectal ulcer (Figure 3). At that point, the patient was asked specifically about rectal prolapse and admitted that she indeed had noted progressive and massive rectal prolapse following defecation over the prior six months.

She subsequently underwent surgery for her symptomatic rectal prolapse with a Ripstein procedure, where a sacral sling was performed using a marlex mesh and sewn to the anterior and side wall of the rectum (4). Her symptoms of mucorrhea subsided shortly after surgery. Follow-up flexible sigmoidoscopy performed six months later showed intact rectal mucosa without findings of ulceration or polypoid lesions.

TABLE 1
Differential diagnosis of solitary rectal ulcer syndrome

Clinical/endoscopic presentation	References
Colitis cystica profunda	7
Idiopathic inflammatory bowel disease	13, 22
Rectal neoplasms	
Adenocarcinoma	23
Benign adenomatous polyps	24
Infectious colitis	12
Human immunodeficiency virus-associated rectal ulcers	25
Drugs	
Oral contraceptives	26
Nonsteroidal anti-inflammatory drugs	27
Ergotamine	28
Colonoscopy bowel preparations	29
Stercoral ulcer	30
Pseudomembranous colitis	13,14
Ischemic colitis	22
Radiation colitis	31
Submucosal lipoma	32

DISCUSSION

SRUS is a benign condition affecting the rectum, which usually presents in women during the third and fourth decades of life, pursuing a chronic course of constipation, mucorrhea, associated rectal prolapse, rectal bleeding and tenesmus (1,2,5). The etiology of this syndrome is thought to be associated with either overt rectal prolapse or internal intussusception, where excessive straining during defecation forces the anterior rectal mucosa downwards against the unyielding pelvic floor, causing trauma and focal ischemia to the mucosa (1-7). Mucosal prolapse with either full thickness rectal prolapse or occult rectal intussusception causes excessive movement of mucosa, and the ischemia that results is due to a combination of traction forces on submucosal vessels, obliteration of submucosal capillaries by fibromuscular proliferation and resulting pressure necrosis of prolapsed mucosa by the anal sphincters (6-9).

The clinical presentation can be variable, as Tjandra et al (1) noted in an earlier retrospective review of 80 patients with SRUS. In their group, the average age at diagnosis was 48.7 years, with a slight female to male predilection with a ratio of 1.4 to 1.0. Seventy-four per cent of patients had nonspecific bowel disturbances consisting of constipation or diarrhea, 56% had rectal bleeding and 26% were asymptomatic. Approximately 25% of patients' conditions were misdiagnosed (Table 1) at the time of initial assessment, proving that this syndrome can be easily missed if specific details in history, course and physical examination are not noted (1,10). Rectal prolapse was associated in 28% of cases.

Mucorrhea occurs in SRUS but is not often the predominant symptom, usually it is overshadowed by tenesmus, constipation, diarrhea, abdominal pain and rectal bleeding. The pathophysiology of mucorrhea in SRUS is unclear, but may be due to altered mucin production by direct stimuli due to ischemia and trauma affecting glycoprotein synthesis and indirectly via cell proliferation (11,12). All mucosal biopsy specimens of the solitary rectal ulcer show the characteristic findings of fibrous obliteration of the lamina propria, disorganization of the muscularis mucosa with extension of muscle fibres into the lamina propria and regenerative crypt epithelium changes with villous configuration and distorted mucosal gland architecture (8-11).

There is an interesting association of polypoid features associated with rectal prolapse, and some authors suggest that this may be a variant of SRUS (13,14). The common features are numerous polyps in the rectosigmoid; signs and symptoms consistent with inflammatory bowel disease, including hematochezia, mucorrhea and tenesmus; negative family history of polyposis or evidence of infection; and histopathology showing minimally inflamed polypoid hyperplastic mucosa with surface erosions and pseudomembranes. This variant of SRUS was described as eroded polypoid hyperplasia, which may be similar to the endoscopic features seen in our case (13,14).

In our case, the multiple polypoid lesions in the rectum were ablated by APC, but severe mucorrhea persisted. It was only during her third follow-up flexible sigmoidoscopy evaluation that we elucidated the history of rectal prolapse, which was then observed on straining. The occurrence of solitary rectal ulcer may represent the evolution from polypoid to ulcerative change in our patient with severe rectal prolapse. However, it is conceivable that APC may have led to a further weakening in the muscularis layer, predisposing the patient to further rectal prolapse and a solitary rectal ulcer (3).

When clinical symptoms have been elucidated and flexible sigmoidoscopy has been performed, other investigations are rarely required and have a limited value in confirming SRUS or rectal prolapse (15). Occasionally a barium enema may show nodularity of the rectal mucosa and thickening of the first valve of Houston, but ulceration is not often shown and in 40% to 50% of cases the study is normal (15,16). Video defecography can be performed if occult prolapse is suspected and may show failure of relaxation of the puborectalis muscle, changes in the anorectal angle and mucosal abnormalities with internal prolapse (15,17). Physiological studies such as anorectal manometry are unlikely to yield significant information on etiology, but reports have demonstrated that concentric needle electromyographic studies of the puborectalis often show failure of relaxation during defecation in 50% of cases with SRUS and may indicate damage to innervation of that muscle (15,18). Overall, previous studies have shown inconsistent findings, and the results of anorectal manometry do not contribute to making the diagnosis or to predicting therapeutic response (15,18). Rectal endoscopic ultrasound may reveal thickening of the muscularis propria and, in some cases, thickening of the internal anal sphincter. The latter is most likely due to abnormal defecation, repeated trauma and ischemia, but this is not a universal finding and associated primary abnormalities must be excluded (15,19). In our case, because the patient was still symptomatic with persistent mucorrhea when the clinical diagnosis of rectal prolapse was made, given the limitations of these other tests, a decision was made to proceed directly to surgery.

The management of this condition is based on the presence of symptoms (15). Usually nonoperative treatment is attempted with bulk laxatives and bowel retraining before considering surgical options for intractable or worsening symptoms. Conservative medical management can include local treatment with application of local steroids, 5-ASA products or sucralfate enemas, but these products have usually been unsuccessful in treating the underlying defecatory disorder, although macroscopic healing did occur (15). Dietary treatment with fibre and bulk-forming agents is often recommended, but there is little evidence to support this and the literature has reported various response rates ranging from 19% to 70% (15,19). Because some patients may have a predominant behavioural disorder of excessive straining with defecation, biofeedback and retraining may be beneficial (15,20). Biofeedback typically includes correction of the pelvic floor defecatory behaviour; regulation of toileting habits; encouragement to stop laxatives, suppositories and enemas; and an attempt to discuss any psychosocial factors that may contribute to their behaviour (15,20). It is often used as an adjunct to surgery and has shown successful results in case series with limited long term follow-up.

In a review by Tjandra et al (1), only 59 of 80 patients had symptoms of SRUS. A limited number (19%) of this subgroup responded to conservative measures alone; 35% of patients had no change in symptoms but these results were not significant enough to warrant surgery, and 27 of 59 (46%) patients underwent surgery for worsening or intractable symptoms. These surgeries included a total of 31 procedures in 27 patients including rectopexy, Ripstien procedure, resection and diversion. Overall, complete symptomatic relief occurred in 20 of the 31 (65%) surgical procedures, with a better response rate noted in patients with polypoid lesions than in those with an ulcerated or inflammatory lesion (1). However, when evaluating surgery for SRUS, Sitzler et al (21) noted that antiprolapse operations resulted in a satisfactory long term outcome (minimum follow-up 12 months) in about 55% to 60% of patients, with results of anterior resection being especially disappointing.

CONCLUSIONS

Eroded polypoid hyperplasia is probably a part of the rectal findings in SRUS. This unusual entity may masquerade as rectal neoplasia, proctitis, colitis cystica profunda and juvenile polyposis. Mucorrhea due to altered mucin production and predominance of sialomucin is a unique clinical feature in this condition. When such lesions are seen, rectal prolapse should be considered. Medical management including biofeedback can be attempted but has had limited success. If symptoms persist, surgical treatment should be undertaken.

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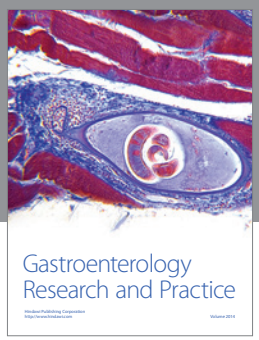

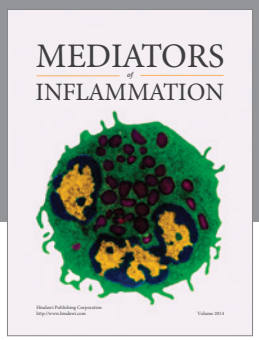

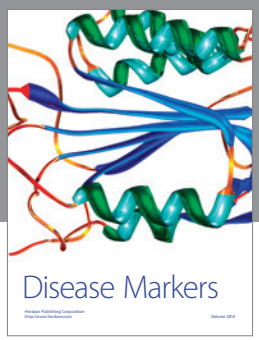

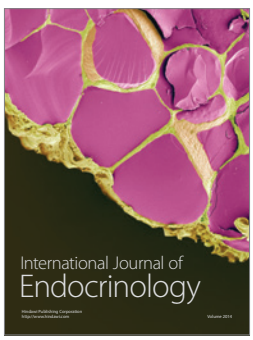

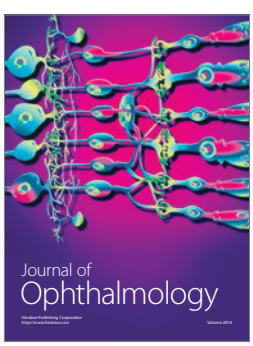

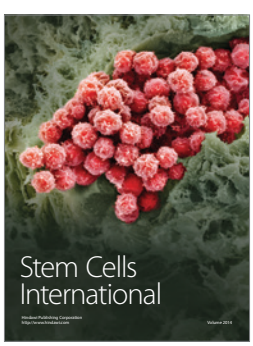

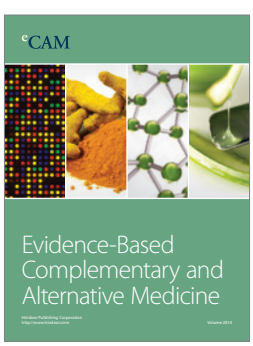

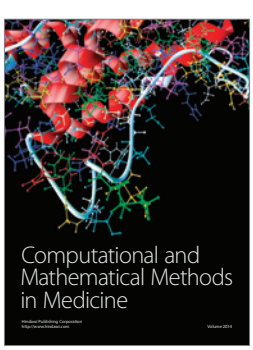

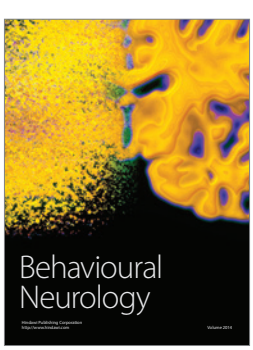

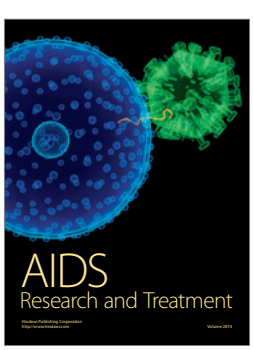

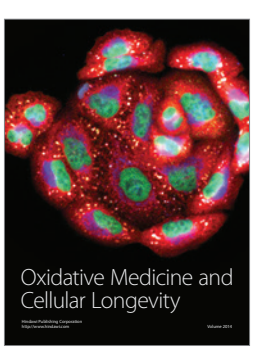

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