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Solitary Rectal Ulcer Syndrome: A Radiologic Diagnosis?

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Abstract. The solitary rectal ulcer syndrome (SRUS) is an uncommon condition in which a solitary area of discrete ulceration is typically found on the anterior wall of the rectum. Between 1981 and 1983, we collected 8 pathologically proven cases of SRUS in which barium enema examinations had been performed (7 double-contrast, 1 single-contrast). Seven patients had rectal bleeding. On the original x-ray report, 4 cases were thought to be normal, but the pathologic tissue had been removed endoscopically in 2 of these cases prior to the radiologic study. The other 4 cases were thought to be abnormal, although the specific diagnosis of SRUS was not suggested in any case. In a blinded rereading of these 8 cases randomly interspersed with 29 other non-SRUS cases, however, the films were interpreted in light of recent radiologic experience with this condition. The same 4 cases were still thought to be normal. In the remaining 4 cases, barium enemas revealed thickened, edematous valves of Houston (3 cases) and a submucosal mass adjacent to the anal verge (1 case). The diagnosis of SRUS was suggested in all 4 cases with only 1 false-positive diagnosis due to a rectal stricture in a patient with endometriosis. Although barium enemas may be normal in patients with SRUS, the presence of thickened, edematous valves of Houston, particularly in a young patient with rectal bleeding, should suggest this condition.

Key words: Colon, radiography – Rectum, ulcer.

The solitary rectal ulcer syndrome (SRUS) is a benign clinical entity in which a solitary area of discrete ulceration is typically found on the anterior wall of the rectum in young patients with rectal bleeding. The diagnosis can be suggested at proctoscopy and subsequently confirmed by classic histopathologic findings on rectal biopsy specimens. However, radiologic studies have not generally been advocated for diagnosing this condition. In 2 articles in the gastroenterological literature, rectal abnormalities were recognized on conventional single-contrast barium enemas in only 2/34 (6%) and 5/35 (14%) cases [1, 2]. As a result, Rutter and Riddell concluded that the barium enema examination usually does not contribute to the diagnosis of SRUS [3].

It is surprising that this entity was not reported in the radiologic literature until 1976, when Lewis et al. described a rectal stricture in a patient with biopsy-proven SRUS [4]. Subsequently, 17 additional cases of SRUS have been documented in the radiologic literature [5–7]. However, these articles were all based on an unblinded, retrospective review of the radiographic findings in biopsy-proven cases. We recently reviewed 8 pathologically proven cases of SRUS seen at our institution between 1981 and 1983, and performed a blinded study to determine whether the diagnosis of SRUS could be suggested radiographically.

Materials and Methods

From 1981 to 1983, 12 pathologically proven cases of SRUS were documented at the Hospital of the University of Pennsylvania. In all cases, the diagnosis was based on classic histopathologic findings on rectal biopsy specimens consisting of fibromuscular obliteration of the lamina propria with thickening and fraying of the muscularis mucosae and frequent extension of smooth muscle and collagen fibers from the muscularis mucosae into the lamina propria (Fig. 1C) [2, 3]. Eight of these 12 patients had barium enema examinations (7 double-contrast, 1 single-contrast). The original x-ray reports were reviewed to determine whether rectal abnormalities had been recognized and whether the possibility of SRUS had been suggested in

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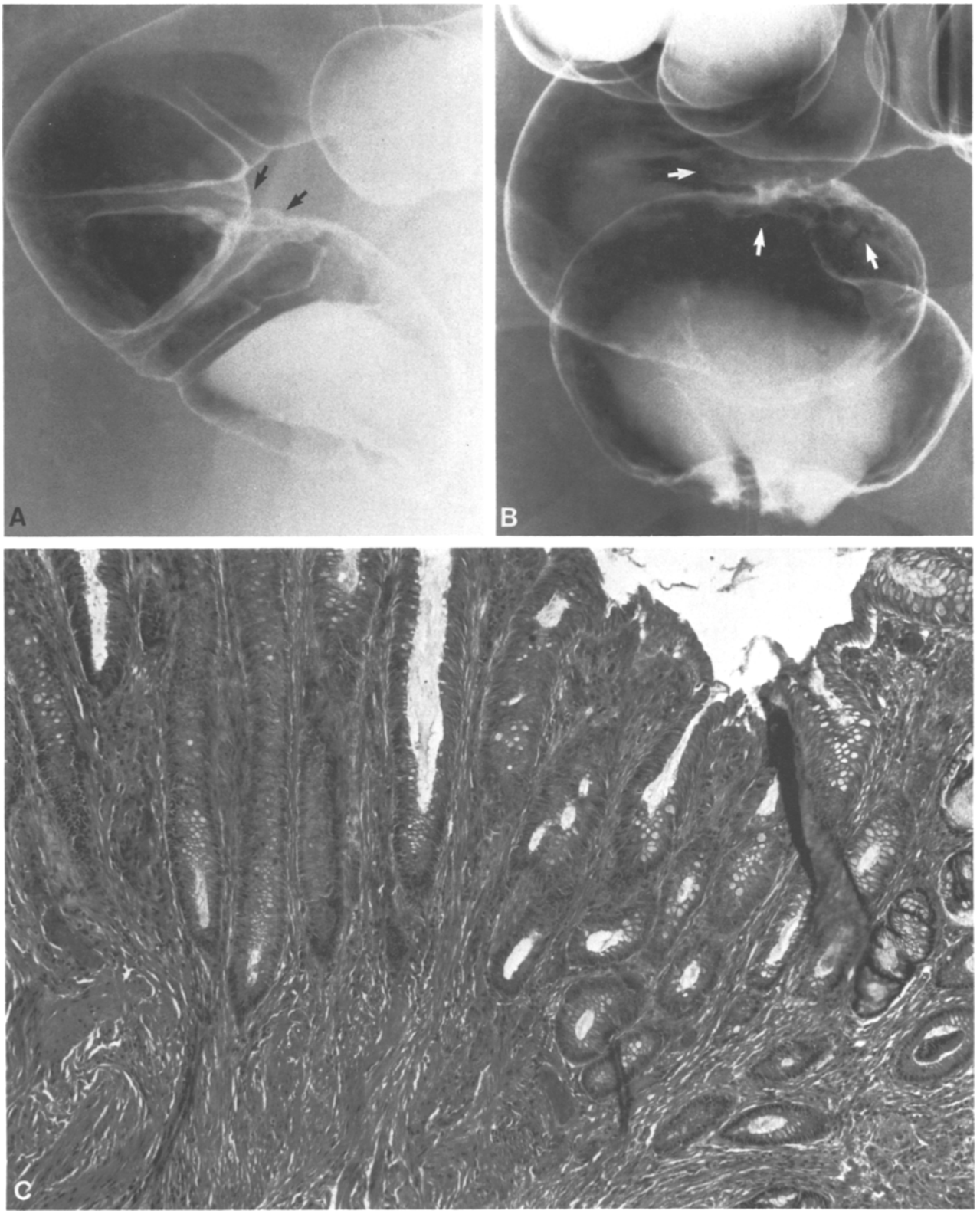


Fig. 1. Solitary rectal ulcer syndrome in 26-year-old woman with rectal bleeding, mucus discharge per rectum, and lower abdominal pain. **A** Lateral radiograph of rectum from double-contrast barium enema shows mild thickening and nodularity of the first 2 valves of Houston anteriorly (*arrows*). **B** Anteroposterior radiograph shows an irregular, nodular mucosa in this region (*arrows*). However, proctoscopy also revealed an anterior rectal ulcer 10 cm from anal verge that could not be seen on the radiologic study. **C** Rectal biopsy specimen shows classic pathologic features of SRUS with fibromuscular obliteration of the lamina propria, thickening of the muscularis mucosae, and crypt hyperplasia (hematoxylin and eosin, 60 ×).

the report. Medical, endoscopic, and pathologic records were also reviewed to determine the clinical findings as well as the proctoscopic and histopathologic findings in these cases.

As a separate part of the study, one of the authors (MSL) performed a blinded rereading of these 8 cases randomly interspersed with 29 other non-SRUS cases in which rectal biopsies had been performed. Thirteen of the non-SRUS patients had a normal rectal biopsy specimen and the remaining 16 patients had diseases other than SRUS, including anal fistulae; ischemic, granulomatous, and ulcerative proctitis; endometriosis; rectal polyps; and rectal carcinoma. Twenty-three of the non-SRUS patients had double-contrast and 6 had single-contrast barium studies. The radiographs were interpreted without knowledge of the history or other pertinent clinical or endoscopic findings. However, the patient's sex and age were given at the time of film review.

The reviewer analyzed all of the radiographs from each barium enema examination and then indicated whether the diagnosis of SRUS was probable, possible, or unlikely in each case. Based on recent radiologic experience with this condition [6, 7], a diagnosis of probable SRUS was made only if the radiographs revealed discrete ulceration on the anterior or anterolateral aspect of the rectum at or near the first valve of Houston. A diagnosis of possible SRUS was made if the radiographs revealed: a rectal stricture; thickened edematous valves of Houston; mucosal nodularity; and/or a submucosal or polypoid mass in the distal rectum adjacent to the anal verge. Finally, SRUS was thought to be unlikely if the radiographs revealed a normal rectum or other evidence of non-SRUS-related disease.

After the initial film review, the cases were unblinded to determine the reviewer's accuracy in diagnosing this condition.

Results

Clinical Findings

In our series of 8 patients, there was an equal sex distribution. The average patient age was 47 years, and the range of ages was 26–72 years. Four patients were below 40 years of age.

Seven of 8 patients (87.5%) had rectal bleeding. In most cases, the degree of bleeding was relatively minor, although 1 patient had major hemorrhage requiring multiple blood transfusions. Other clinical findings included mucus discharge (4 cases), altered bowel habits with constipation and/or diarrhea (4 cases), anorectal or lower abdominal pain (3 cases), rectal prolapse (2 cases), and tenesmus (1 case). One patient was asymptomatic. The average duration of symptoms prior to the diagnosis of SRUS was 6.3 years with a range of 1–11 years.

Endoscopic Findings

Proctosigmoidoscopy revealed a single area of discrete ulceration on the anterior wall of the rectum in 2 cases and on the posterior wall in 1 case. The ulcers were all located 8–10 cm from the anal verge. Both anterior ulcers were associated with thickened, nodular rectal folds at the level of ulceration. In a fourth case, multiple areas of punctate ulceration were associated with generalized erythema and circumferentially thickened, distorted rectal folds 8–11 cm from the anal verge. While these ulcers were all thought to have a benign appearance, a specific diagnosis of SRUS was suggested proctoscopically in only 1 case.

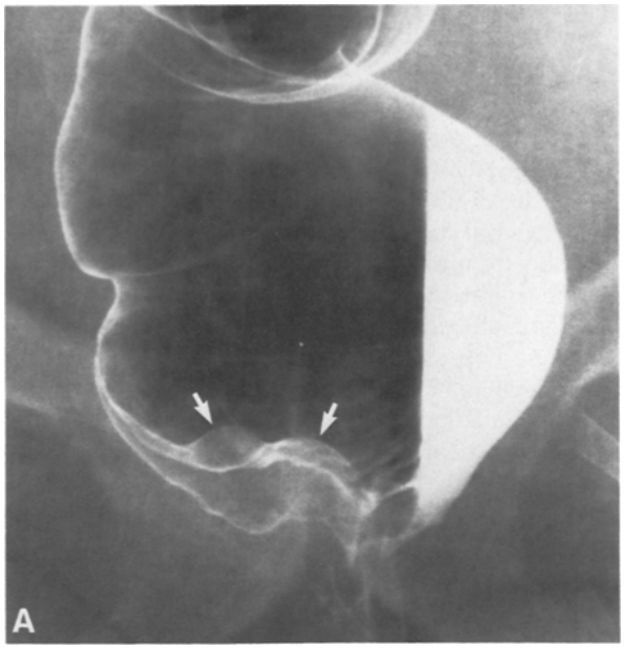
In the remaining 4 cases, proctosigmoidoscopy revealed polypoid lesions in the rectum without evidence of ulceration. In 2 cases, the only proctoscopic finding was a single polyp smaller than 1 cm in the distal rectum. In both of these cases, endoscopic polypectomy was performed prior to the barium enema examination. In a third case, a mass of unspecified size adjacent to the anal verge was thought to represent a chronically inflamed anal cyst. In the final case, the submucosal appearance of a 1-cm mass on the right lateral rectal wall just above the anal verge was mistaken for a thrombosed internal hemorrhoid. Thus, a diagnosis of SRUS was not suggested proctoscopically in any of these 4 cases.

Histopathologic Findings

Rectal biopsy specimens revealed characteristic histopathologic findings of SRUS in all cases (Fig. 1C). Mucosal ulceration was present in 6/8 or 75% of cases, but these ulcers were almost always superficial in nature. In 2 cases, biopsy specimens also revealed histologic findings of localized colitis cystica profunda with displaced mucosal glands in the submucosa and, as a result, pools of mucin trapped beneath the muscularis mucosae (Fig. 2B). In 1 of these cases preliminary frozen sections were interpreted as showing possible mucinous adenocarcinoma of the rectum, but examination of the final pathologic specimen revealed SRUS with localized colitis cystica profunda.

Radiographic Findings

Original Film Reading. On the original x-ray report for the barium enema examinations, the rectum was thought to be normal in 4 cases (3 double-contrast, 1 single-contrast). In the 4 abnormal cases (all double-contrast), the reporting radiologist described thickened, nodular valves of Houston and/or rectal folds (3 cases) (Figs. 1, 3, 4) and a slightly lobulated submucosal mass in the distal rectum adjacent to the anal verge (1 case) (Fig. 2A). The radiologic differential diagnoses were proctitis or rectal varices (1 case), endometriosis or metastases (1 case), primary or metastatic carcinoma (1 case), and primary carcinoma or unusually prominent internal hemorrhoids (1 case). However, the specific diagnosis of SRUS was not suggested in any case.



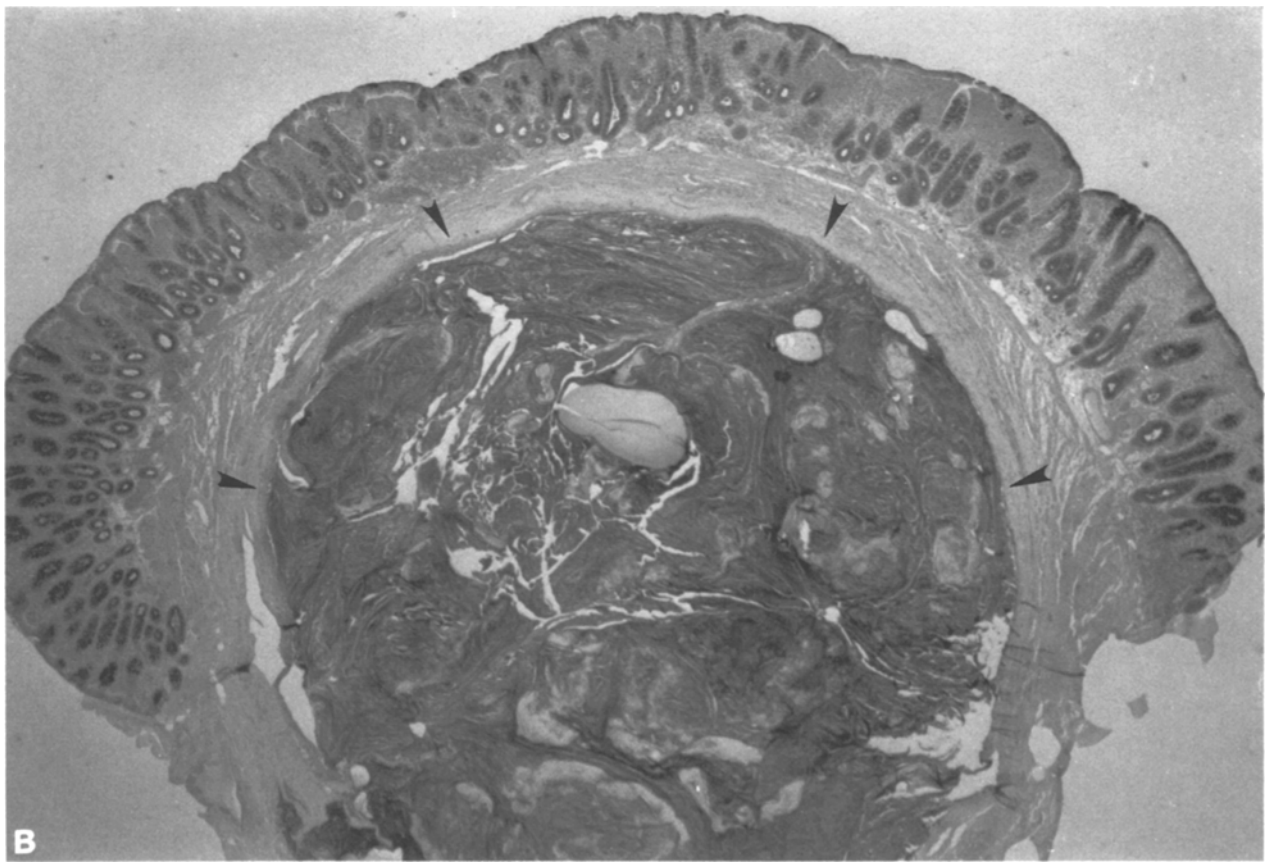


Fig. 2. An asymptomatic 72-year-old man. **A** Left lateral decubitus radiograph from double-contrast barium enema shows a slightly lobulated submucosal-appearing mass in distal rectum adjacent to anal verge (*arrows*). **B** Histologic section from excisional biopsy specimen shows a mucus-filled epithelial-lined cyst (*arrowheads*) in the submucosa (i.e., localized colitis cystica profunda) (mucicarmine, 20×). Higher magnification also revealed classic features of SRUS.

Blinded Rereading. In a blinded rereading of these 8 cases randomly interspersed with 29 other non-SRUS cases, SRUS was thought to be probable in no cases, possible in 5 cases, and unlikely in 32 cases (17 had a normal rectum and 15 had diseases other than SRUS). The possibility of SRUS was suggested radiographically in 5 cases by the presence of thickened, edematous valves of Houston, predominantly involving the first valve (3 cases) (Figs. 1, 3, 4), a slightly lobulated submuco-

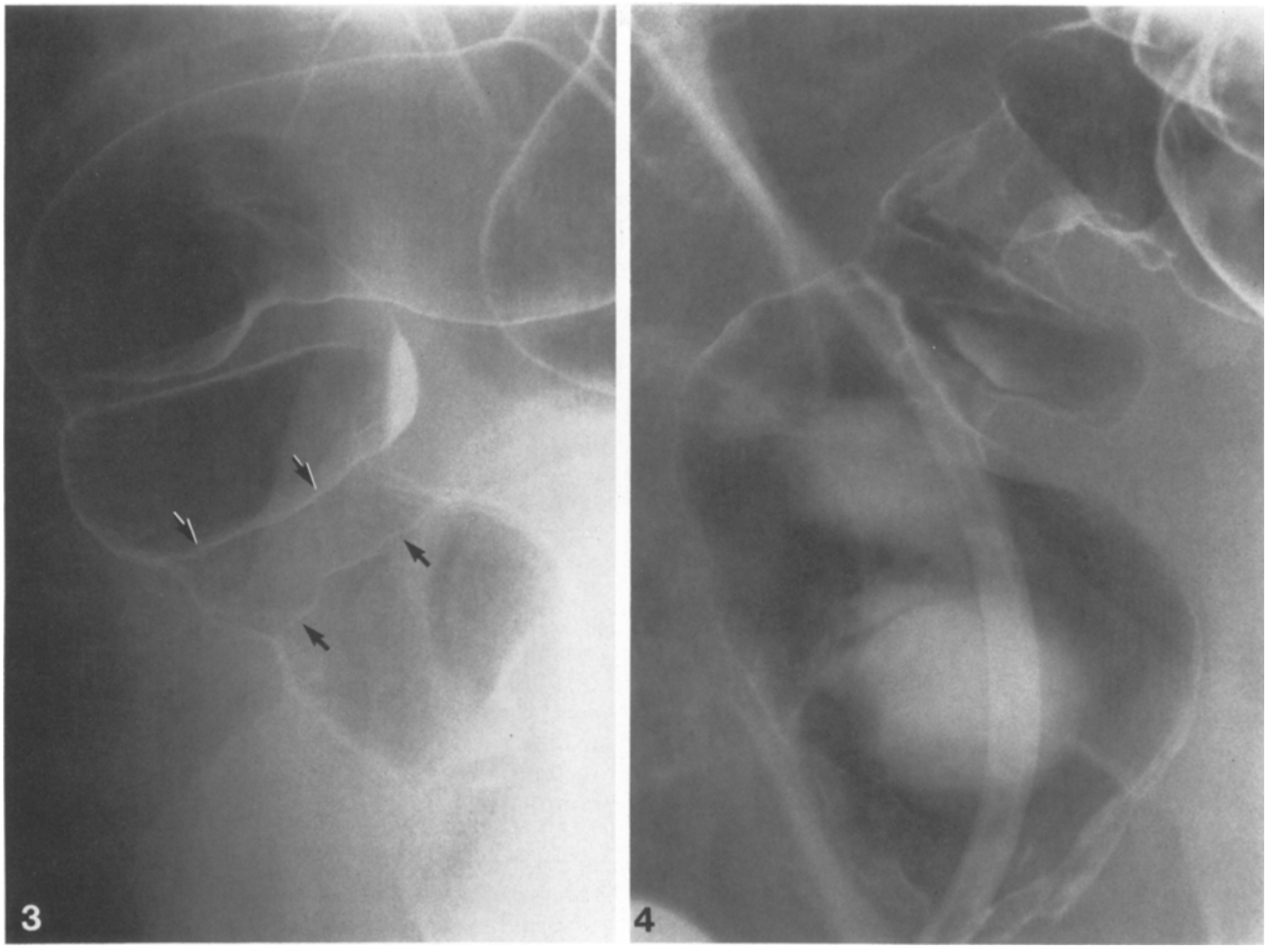


Fig. 3. A 33-year-old man with rectal bleeding. Lateral radiograph of rectum shows a markedly thickened, edematous first valve of Houston (*arrows*) without ulceration. This appearance seems to be characteristic of SRUS on the double-contrast examination. At proctoscopy, however, a shallow ulcer was also detected anteriorly 8 cm from the anal verge.

Fig. 4. A 57-year-old woman with rectal bleeding and intermittent left lower quadrant abdominal pain. Steep oblique radiograph of rectum shows markedly thickened valves of Houston with circumferential narrowing at level of second valve or rectosigmoid junction. (Note air insufflation catheter in rectum.) Proctoscopy confirmed the presence of thickened rectal folds but also revealed multiple superficial ulcerations not seen radiographically in this region.

sal mass adjacent to the anal verge (1 case) (Fig. 2A), and a rectal stricture (1 case). Four of these 5 patients had biopsy-proven SRUS with a single false-positive radiologic diagnosis due to a rectal stricture in a patient with endometriosis. Thus, the diagnosis of SRUS was suggested radiographically in 4/8 patients, although discrete ulcers were not detected in any case. In the remaining 4 patients with SRUS, barium enema examinations were normal. However, it should be noted that the pathologic tissue had been removed endoscopically in 2 cases prior to the radiologic study. Thus, barium enemas were abnormal in 4/6 or 67% of the unaltered cases of SRUS.

Discussion

The solitary rectal ulcer syndrome (SRUS) is an uncommon condition that classically presents with a persistent, nonhealing ulcer on the anterior aspect of the rectum. However, the name of the condition is misleading, since there can also be multiple ulcers or a localized proctitis without ulceration [1–3]. The exact pathogenesis of SRUS is uncertain, although it is often associated with rectal prolapse [8–11] or a history of straining at stool [3]. Electromyographic studies have shown that many patients with SRUS have pelvic muscle discoordination during defecation with persistent contraction

of the puborectalis muscle [12]. As a result, it has been postulated that repeated straining causes occult prolapse of the anterior wall of the rectum with ulceration of the prolapsed mucosa due to mechanical trauma and/or ischemia [1, 3, 13]. This theory would explain why ulceration is typically found on the anterior rectal wall.

This syndrome usually occurs in patients under the age of 40 [1–3], but older individuals may occasionally be affected. There is an approximately equal sex distribution [2, 3] or perhaps a slight female predominance [1, 8]. Most patients present clinically with mild rectal bleeding [1–3, 8, 11], although major gastrointestinal blood loss requiring multiple transfusions has been reported [2]. Unfortunately, the clinical findings are nonspecific, and the average interval between the onset of symptoms and diagnosis is 5 years [1]. This syndrome may be treated with a high-roughage diet, steroid enemas, sulfasalazine, and local excision [1], but medical and surgical methods of treatment are often unsatisfactory [2, 8, 14].

Solitary rectal ulcer syndrome can be diagnosed on proctoscopy by the typical finding of a solitary, benign-appearing ulcer with discrete, slightly raised margins on the anterior or anterolateral wall of the rectum 7–10 cm from the anal verge [2, 3, 11]. However, these ulcers may be multiple in 25–30% of cases [2, 8], they occasionally may be found on the posterior or lateral wall of the rectum [2], and their location may vary between 3 and 15 cm from the anal verge [2, 8, 11]. Other patients may have a localized proctitis with a nodular, lumpy, or erythematous mucosa but no discrete ulcers [1–3, 8]. This appearance has been described as the "preulcerative" phase of SRUS, on the assumption that ulceration invariably occurs [3]. However, long-term follow-up in these cases has rarely documented the development of ulcers [1, 8], so that it could be more aptly described as a "nonulcerative" variant of this disease.

Because SRUS is an uncommon condition with a variety of endoscopic findings, the diagnosis is usually made based on findings on rectal biopsy specimens rather than the gross appearance at proctoscopy. In our study, SRUS was suspected endoscopically in only 1/8 cases (12.5%), but rectal biopsy specimens revealed classic histopathologic findings in all cases (Fig. 1C) [2, 3]. Thus, it should be recognized that a definitive diagnosis of SRUS can be made on histologic criteria, regardless of the presence or absence of ulceration.

In 2 of our cases, rectal biopsy specimens also revealed localized colitis cystica profunda with mucus-filled epithelial cysts in the submucosa (Fig. 2B). Others have previously reported an association between SRUS and localized colitis cystica profunda [2, 15, 16]. It has been postulated that the latter condition develops as a complication of SRUS due to extension of regenerating surface epithelium into the submucosa [3, 16, 17]. While colitis cystica profunda is a benign condition, pathologists have occasionally mistaken these mucus-filled cysts for invasive mucinous adenocarcinoma [2, 15, 18]. Indeed, a preliminary diagnosis of rectal cancer was made in 1 of our cases. Thus, pathologists should be aware of the association between SRUS and localized colitis cystica profunda, so that unnecessary radical surgery can be avoided in these patients.

The presence of a solitary, benign-appearing ulcer on the anterior wall of the rectum near the first valve of Houston on barium enema examination should suggest the diagnosis of SRUS [5]. Even when present, however, these ulcers are relatively shallow and can easily be missed on conventional single-contrast barium studies. Of 18 cases of SRUS reported in the radiologic literature, ulceration was detected radiographically in only 6 (33%) with discrete anterior rectal ulcers found in only 4 (22%) [4–7]. Even with careful double-contrast technique, Feczko et al. were able to demonstrate ulcers in only 2/6 cases with ulceration at proctoscopy [6]. In our study, we failed to detect ulcers radiographically in any of 4 cases with proctoscopic evidence of ulceration. Thus, most cases of SRUS will be missed on barium enema examination if anterior rectal ulceration is the only radiologic criterion for diagnosing this condition.

However, it is now recognized that SRUS may be manifested by a spectrum of radiographic findings, including nodularity of the rectal mucosa, thickened valves of Houston, ulceration, strictures, and submucosal or polypoid rectal masses [6, 7]. Feczko et al. most commonly observed a nodular rectal mucosa and/or thickening of the first valve of Houston on double-contrast examinations [6]. In our study, 3/4 patients with abnormalities in the rectum had thickened, edematous valves of Houston without discrete ulcers (Figs. 1, 3, 4). In a fourth patient with SRUS and localized colitis cystica profunda, the barium enema examination revealed a slightly lobulated submucosal mass adjacent to the anal verge (Fig. 2A). However, we were able to suggest the diagnosis of SRUS radiographically in all 4 abnormal cases on blinded rereading of the films. We therefore believe that the presence of thickened, nodular valves of Houston on barium enema examination should suggest this condition.

While SRUS may be suspected on barium studies, a variety of conditions should be considered in the differential diagnosis. Rectal ulceration may be found in other types of proctitis (including granulomatous or ulcerative, venereal, and traumatic proctitis) or in ulcerated tumors [5–7]. Thickened, nodular valves of Houston can also result from other types of proctitis or circumferential infiltration by endometriosis or metastases [7]. Rectal strictures may be caused by numerous conditions, including granulomatous or ulcerative colitis, endometriosis, lymphogranuloma venereum, ischemia, radiation, and primary or metastatic tumors [4–7]. Finally, a submucosal or polypoid mass in the distal rectum may result from benign or malignant tumors or unusually prominent internal hemorrhoids [14]. However, the clinical history is often helpful in differentiating these conditions.

In summary, our experience suggests that barium enema examinations may be normal in patients with SRUS. In abnormal cases, discrete ulcers are rarely seen radiographically, but the presence of thickened, edematous valves of Houston, particularly in a young patient with rectal bleeding, should suggest this condition. In such cases, the proctoscopist and pathologist should be alerted about the radiologic findings, so that endoscopic biopsy specimens can be obtained for a definitive diagnosis.

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