Solitary Rectal Ulcer Syndrome : Two Case Reports†

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Abstract

Owing to its rarity, solitary rectal ulcer syndrome (SRUS) is often misdiagnosed as malignant ulcer, or ulcer in association with inflammatory bowel disease. We present two adult females with anorectal symptoms (i.e. pain, tenesmus and bowel habit changes). Both had normal levels of serum carcinoembryonic antigen. Barium enema revealed irregular mucosa with stricture of the lower rectum. An ulcer, 2.7 cm in diameter, was found in one patient but not the other. Rectal biopsy under sigmoidoscopy demonstrated non-specific inflammation, without evidence of malignancy. Because of the intractable symptoms and the inability to discriminate between malignant and benign conditions, exploratory laparotomy was performed, followed by low anterior resection of the rectum. Histological examination of both specimens showed submucosal rectal fibrosis with a non-specific ulceration in one. These findings were compatible with SRUS. The patients' symptoms improved dramatically after the resection and they remain well, five months and one year after surgery. Awareness of this rare anorectal condition is necessary for appropriate management particularly to avoid unnecessary abdomino-perineal resection.

Key word : Solitary Rectal Ulcer Syndrome, Case Report

Solitary Rectal Ulcer Syndrome (SRUS), an uncommon benign rectal disease, was first described as early as the 19th century by Cruveilhier[1], but was not widely recognized until the 1969 publication of Madigan and Morson[2]. Most patients have rectal bleeding and a disturbance of bowel function[2,3] which may or may not be associated with ulcer. The ulcerative lesions are mostly located at the anterior or anterolateral wall of the rectum [2-4]. The characteristic histological changes in the

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rectal biopsy specimens are fibromuscular obliteration of the lamina propria accompanied by the thickening of the muscularis mucosae[2,5]. The cause of the condition is unknown, but it has been ascribed to incomplete or total rectal prolapse[5,6]. Various treatment strategies have been advocated, ranging from conservative management to a variety of surgical procedures. The optimal treatment for the condition remains unclear[3].

We report two cases, which resemble rectal carcinoma in clinical presentation, endoscopic appearance, and radiographic appearance, to remind surgeons of this uncommon rectal condition and avoid unnecessary abdomino-perineal operation.

CASE REPORT

Case 1:

A 45-year-old female had anal pain and weight loss (5 kg) for one month before being seen in hospital. Physical examination was normal except that a rectal examination revealed a constrictive lesion in her rectum. She had a normal level of serum carcinoembryonic antigen. Barium enema showed an irregular mucosa with stricture of the lower rectum (Fig. 1). The initial radiological diagnosis was rectal carcinoma. Sigmoidoscopy revealed a multiple polypoid mass with white plaque, 5 cm from the anal verge. The rectal biopsy under sigmoidoscopy revealed chronic inflammation. No malignant cells were observed. Because of the intractable symptoms and the inability to discriminate between benign and malignant conditions, exploratory laparotomy with tumor resection and end-to-end anastomosis was performed. Pathological examination of the resection specimen (S.41-01882) revealed no ulceration of the mucosal surface of the whole rectal segment which was 10 cm long. Histologically, H&E and Masson trichrome stains showed diffuse obliteration of the lamina propria of the mucous membrane by proliferation of fibroblasts producing abundant collagen fibers admixed with smooth muscle fibers derived from the relatively thick muscularis mucosae (Fig. 2A & 2B). The presence of smooth muscle fibers was better visualized by 1A4 stain (an immunoperoxidase stain for smooth muscle actin). The submucosa of the rectum showed marked fibrosis. The findings, when rectal prolapse was excluded, were highly suggestive of solitary rectal ulcer. The patient is now, six months after treatment, gaining weight and without anal pain.

Fig. 1. Barium enema shows an irregular mucosa with stricture of the lower rectum.

Case 2:

A 34-year-old female presented with a 2-month history of mucous bloody stools and change of bowel habits. The rectal examination showed rectal ulcer anteriorly. She had a normal level of carcinoembryonic antigen. Barium enema revealed an ulcer with a constrictive lesion of the lower rectum, being diagnosed as carcinoma of the rectum (Fig. 3). Sigmoidoscopy revealed a 2.7 cm in diameter, punched-out ulcer 8 cm from the anal verge on the anterior rectal wall. Rectal biopsy under sigmoidoscopy revealed chronic proctitis with no evidence of malignant cells. For the same reason as the first case, low anterior resection with end-to-end anastomosis was performed. The resected segment of the rectum (S.40-5751) was 8.5 cm long and showed a large flat ulcer of 2.7 cm in diameter in the anterior wall. Microscopic examination disclosed similar findings to those in the first case except that mucosal ulceration was present (Fig. 4). The patient is now, one year after operation, gaining weight, completely asymptomatic and had normal finding on sigmoidoscopy.

DISCUSSION

SRUS has been underdiagnosed in Thailand because it is an unfamiliar condition. It is characterized by a combination of symptoms, clinical findings and typical histological abnormalities.

Fig. 2A. Photomicrography of S41-01882 (case 1) shows obliteration of the lamina propria and decreased globlet cells of the epithelial tubules. (H&E, x 200).

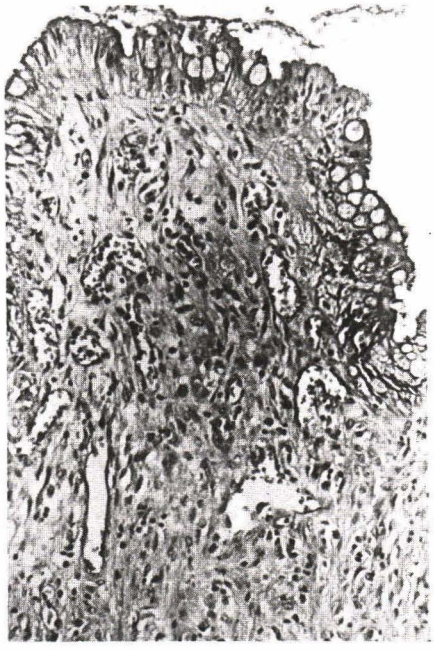

Fig. 2B. Photomicrograph of S41-01882 (case1) illustrates nonulcerated rectal mucosa with thick muscularis mucosae and proliferation of fibroblasts producing collagen fibers admixed with smooth muscle fibers. (H&E, x 100).

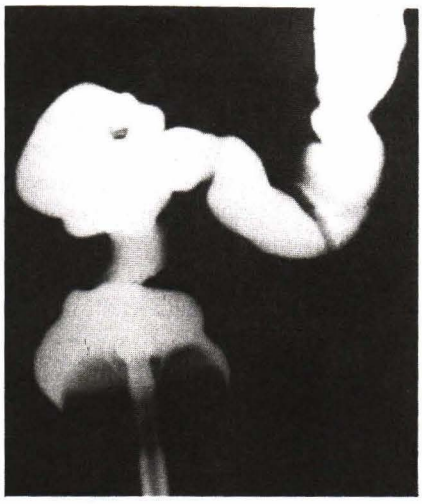

Fig. 3. Barium enema reveals an ulcer with a constrictive lesion of the lower rectum.

Fig. 4. Photomicrograph of S41-5751 (case 2) shows ulceration of the rectal mucosa. The adjacent mucosa shows obliteration of the lamina propria due to replacement of normal tissue by proliferative collagen fibers and smooth muscle cells and thick underlying muscularis mucosa. (H&E, x 40).

count revealed a hematocrit of 31 per cent, WBC 10,000 cell per mm^3, neutrophils 66 per cent lymphocyte 27 per cent, monocytes 5 per cent, eos 1 per cent. The serum electrolyte showed Na^+ 135 mmole/1, K^+ 4.4 mmole/1, CI 17 mmole/1, and CO_2 24 mmole/1. The serum albumin was 25.9 g/dl. The upper GI study revealed narrowing of the mid and lower esophagus with minimal mucosal irregularity. The pylorus showed almost complete obstruction due to fibrosis, The gastroscope showed a shallow ulcer and mild luminal narrowing of the esophagus, fibrosis and obstruction at the pylorus and antrum of the stomach. Subtotal gastrectomy and Billroth-II anastomosis were performed. The pathological findings were similar to those of the two previous cases. The patient was discharged 7 days after surgery without complication. He was seen one month later and was noted to have no problem in ingestion of food.

DISCUSSION

Accidental or intentional ingestion of caustic substances results in burning of various degree in the esophagus, stomach, or proximal small bowel. It may present acutely as perforation or necrosis, necessitating emergency surgery, or it may manifest later owing to progressive scarring. Acids and alkalines affect tissue in a different manner. Alkalines dissolve tissue and therefore penetrate more deeply, while acids cause coagulative necrosis which limits their penetration[2]. Vixol is one of the most common household products used for cleaning toilet bowls. It is composed of 23 per cent hydrochloric acid, 3.5 per cent alkylphenol polyglycol ether, and 2.5 per cent of acrylic copolymer anhydrous. We reviewed three cases of gastric damage following ingestion of Vixol. We thought that the main pathological finding was due to hydrochloric acid which is a corrosive substance. Gastric outlet obstruction was the late sequelae of all presented cases. The main pathology occurred at the prepyloric region along the lesser curvatures. A significant esophageal lesion was found in the third case who ingested a large amount of this agent (960 ml), whereas, the other two cases who ingested a lesser amount showed minimal changes. Chaudhar et al[3] reported 34 cases of corrosive induced gastric injury similar to ours. However, all of those cases showed extensive perigastric adhesion which was not found in our cases. In Chaudhar's study, almost all cases had preoperative jejunostomy to improve nutritional status and elective surgery was done 3 to 24 months (average 7 months) later. Our cases were operated on earlier at 1,2,4 months respectively. In spite of earlier elective surgery without preoperative jejunostomy we did not find more postoperative complications. Although the ingestion of strong acids may cause injury to any part of the upper gastrointestinal tract, squamous epithelium seems to be more impervious to its effect than glandular mucosa. A

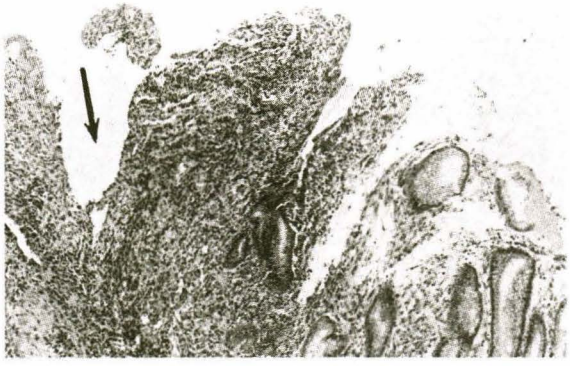

Fig. 1. (H & E ; original magnification x 50) The photomicrograph shows inflammed mucosa and presence of ulcer (arrows) in the stomach.

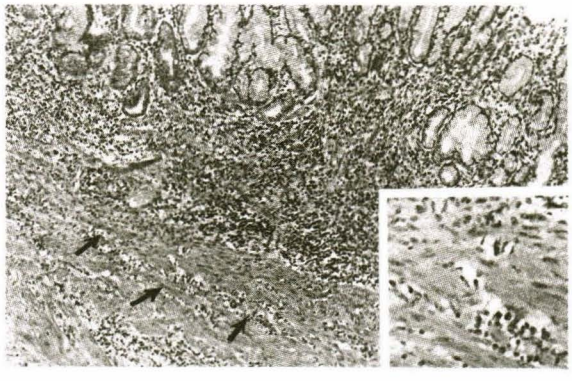

Fig. 2. (H & E ; original magnification x 10) The photomicrograph shows moderate infiltration of inflammatory cells in the mucosa with thickening and derangement of muscalaris mucosa (arrows) in the stomach. Inset : (H & E ; original magnification x 100) The thickened muscularis mucosa shows hyperplasia of smooth muscle fibers and infiltrated with inflammatory cells.

Rectal bleeding, disordered defecation and anal pain are associated with a benign rectal lesion with characteristic histological appearance[3]. The lesions are not necessarily solitary. The macroscopic findings range from hyperemia to ulceration or even a polypoid lesion[7]. Rectal prolapse must be excluded since this can produce similar histological changes.

The most common clinicopathologic misdiagnoses in SRUS patients with rectal ulcers or mucosal hyperemia include Crohn's disease and mucosal ulcerative colitis. In patients with "polypoid" SRUS, diagnostic confusion is usually with a neoplastic polyp[7].

Sometimes the clinical and radiologic findings can mimic rectal carcinoma and, conversely, underlying malignancy, metastatic carcinoma from stomach or ovary, direct invasion of anal squamous cell carcinoma or prostatic adenocarcinoma, can produce characteristics of SRUS-like histopathology[8]. Therefore, if SRUS is suspected in any patient with anorectal symptoms, a tissue biopsy is recommended to exclude malignant disease and to confirm the diagnosis of this benign condition.

As the real pathogenesis of this condition remains unknown, the definitive treatment has not been established. Conservative treatments, including laxatives, fiber supplementation and attempted reduction of straining, are of no proven benefit and there are no topical agents known to improve this condition[9]. In one prospective study the use of biofeedback retaining was reported to improve symptoms for SRUS in a significant number of patients, but the median follow up period was only nine months. The authors concluded that biofeedback retraining is a useful treatment and long term studies are required[10]. Other non-invasive treatments, e.g. human fibrin sealant[11] and NdYAG lasers [12], have also been suggested recently but their long-term benefit is uncertain.

The optimal surgical procedure is still unknown, but local excision, rectopexy, and diversion have been tried with variable results[2,3]. Thus, surgical treatment may be useful if medical treatment fails. When rectal prolapse is present, a rectopexy would be the most favorable procedure for abnormal anorectal symptoms[13]. Our suggestion for SRUS is to remove the diseased segment by means of low anterior resection whenever possible. The main reasons are not only to cure the symptoms but also to rule out malignancy with certainty.

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กลุ่มอาการแผลเดี่ยวในลำไส้ใหญ่ส่วนปลาย : รายงานผู้ป่วย 2 ราย

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ภาวะกลุ่มอาการแผลเดี่ยวในลำไส้ใหญ่ส่วนปลายพบได้ไม่บ่อย และบางครั้งได้รับการวินิจฉัยผิดเป็นเนื้อร้ายของลำไส้ใหญ่ส่วนปลาย หรือโรคของทางเดินอาหารอักเสบ รายงานผู้ป่วยหญิง 2 ราย ที่มาพบแพทย์ด้วยอาการปวดทวารหนักปวดขณะถ่ายอุจจาระและมีการเปลี่ยนแปลงลักษณะของการขับถ่ายอุจจาระ การตรวจวินิจฉัยด้วยการสวนแป้งทางทวารหนักเข้าได้กับลักษณะเนื้อร้ายของลำไส้ใหญ่ส่วนปลาย แต่ผลการตรวจชิ้นเนื้อที่ได้จากการส่องกล้องทางทวารหนักไม่พบลักษณะของเนื้อร้าย เนื่องจากไม่สามารถวินิจฉัยแยกโรคเนื้อร้ายของลำไส้ใหญ่ส่วนปลาย ผู้ป่วยจึงได้รับการรักษาด้วยวิธีการผ่าตัดโดยการตัดลำไส้เฉพาะส่วนที่มีพยาธิสภาพ ผลการตรวจทางพยาธิพบลักษณะเฉพาะที่เข้าได้กับกลุ่มอาการแผลเดี่ยวของลำไส้ใหญ่ส่วนปลาย และผู้ป่วยมีอาการดีขึ้นอย่างชัดเจนภายหลังจากการผ่าตัด

การตระหนักถึงกลุ่มอาการแผลเดี่ยวในลำไส้ใหญ่ส่วนปลายที่พบได้ไม่บ่อยนี้จึงจำเป็นเพื่อหลีกเลี่ยงการผ่าตัดด้วยวิธีการตัดลำไส้ใหญ่ส่วนปลายและทวารหนักโดยไม่จำเป็น

คำสำคัญ : กลุ่มอาการแผลเดี่ยวในลำไส้ใหญ่ส่วนปลาย, รายงานผู้ป่วย

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