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Case Report

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SOLITARY RECTAL ULCER MIMIC RECTAL CANCER

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Abstract

Solitary rectal ulcer (SRU) is rare benign disease of poorly understood etiology. The term solitary ulcer is misnomer because the lesion is solitary in only 20% of patients and ulcer present in 40% of the patients only. The lesion has various sizes and shapes that ranges from mucosal erythema to single or multiple ulcers to ulcer-nodular lesion or could be a polypoid or fungating mass. The estimated incidence of solitary rectal ulcer is about 1:100000 annually. Males and females are affected equally with slight predominance toward female patients. The disease can affect children and elderly patients also, although rare. The disease involves straining during defecation, a sense of incomplete evacuation, digital evacuation of the rectum, rectal prolapse and occasionally passing of blood and mucus by rectum. Clinical awareness confirmed by flexible sigmoidoscopy and biopsy are main diagnostic tools. Furthermore, the colonoscopic findings may mimic that of rectal cancer and inflammatory bowel disease and the histopathological results are inadequate due to fibrotic rectal mucosa. Treatment of solitary rectal ulcer depends on the severity of the condition with conservative measures, bowel regimen, and change of life style for mild cases and surgical procedure for sever refractory cases.

Case study: A 17 year old female patient presented with complaints of straining and rectal bleeding while defecation of 7 months duration, chronic and long-term history of constipation with usual digital evacuation of her rectum at the end of her bowel motion. She was treated first conservatively as a case of chronic anal fissure after consulting a general surgeon but no improvement noticed was in her condition for which sigmoidoscopy was planned. The sigmoidoscopic findings revealed evidence of circumferential annualr ulcer with polypoid nodular mass about 4 to 5 cm from anal verge. Multiple biopsies was taken from the lesion and sent for histopathology and diagnosis of well-differentiated adenocarcinoma of the rectum was the diagnosis. Surgical resection in form of low anterior resection

or abdomino-perineal resection with adjuvant chemotherapy and radiotherapy were advised. Patient's parents refused surgery and preferred to review her condition by other specialist to confirm the diagnosis. Patient was seen later by a gastro-enterologist surgeons team in highly specialized gastroenterology center who re-evaluate the patients by reviewing her initial paraffin block and slides and the result was that there was no evidence of dysplasia or invasive malignancy. Pelvic MRI revealed thickening of the rectum with luminal compromise with surrounding diffuse edema. PET CT scan were done also which revealed metabolically active rectal Sigmoidoscopy was repeated growth. which showed polypodal growth 4 cm from anal verge from which multiple pieces was taken for histopathology and the result was benign lesion consistent with solitary rectal ulcer with no any evidence of dysplasia or malignancy. Rectal endoscopic ultrasound was done which showed an ulcero-nodular lesion at 4-5, thickened submucosa and thickened internal sphincter with few small perirectal lymph nodes seen. Explorative laparoscopy was done and showed redundant sigmoid colon was seen intrawhich operatively for laparoscopic sigmoidectomy and rectopexy was done. Postoperative period was uneventful. Patient discharged from hospital in the fifth postoperative day on soft diet after passing her first bowel motion.

Discussion

Solitary rectal ulcer (SRU) is a rare benign and poorly understood rectal disorder. It affect both sexes equally with slight female predominance. The incidence of solitary rectal ulcer is uncertain or could be underestimated. It has been estimated in one study to be about 1: 100000 patient per year. The average age at diagnosis is 48 years with a range from 15 to 75 years¹.

Solitary rectal ulcer is best described as "the three lies disorder" because the lesion is not always solitary and not always confined to the rectum and not always ulcerative². It is chronic benign disease characterized by diverse and non specific clinical and endoscopic features which are close to or similar to other diseases such as rectal cancer or inflammatory bowel disease. It is manifested mainly as bleeding per rectum with or without mucus discharge, sense of incomplete evacuation, tenesmus, chronic constipation. Other less common symptoms include perianal and pelvic pain, diarrhea and rectal prolapsed or even asymptomatic in about 20% of cases^{2,3}.

Rectal bleeding which ranges from mild to severe hemorrhage is present in almost all cases of SRU. Massive blood transfusion necessitate sometimes urgent blood transfusion and sigmoidoscopy or colonoscopy. Rectal bleeding, tenesmus, long-term and chronic constipation and rectal digitations are the main clinical feature of our case^{4,5}.

The exact etiology of this disorder is still understood, poorly Several factors appeared to contribute to etiology of SRU. The two factors, however that could involve in the development of SRU are direct trauma of the rectum and reduced blood flow (ischemia) to the area which is commonly the anterior rectal wall. The main cause of such trauma and ischemia is the rectal prolapsed that developed due excessive and prolonged straining during defecation especially when it accompanied by contraction of puborectalis which encircles the anterior portion of upper part of anal canal. The combined effects of downward pressure and movement of mucus membrane together with stool during defecation and upward pressure due to contraction of puborectalis muscle may create enough pressure that results in injury and ischemia of rectum^{1,2,4}. Other factors that may injure rectal mucosa and subsequently initiate SRU chronic constipation and hard impacted stool, straining during defecation to evacuate the impacted stool by fingers. Rectal prolapse, chronic constipation and excessive straining, and rectal digitations are all present in our case.

The most common colonoscopic findings of SRU is the presence of ulcer on the anterior rectal wall in about two third of cases. The ulcer could be single, multiple, or circumferential about 4 or 5 cm from anal verge but it could be as high as 20 cm from anal orifice. The size of the ulcer is usually less than 2 cm with size ranges from 0.5 to 5 cm. The ulcer is usually superficial and non penetrating. It bleeds easily but rarely produces massive rectal bleeding. Thickening of the rectal wall mainly the muscularis propria is a common finding in SRU as shown by rectal ultrasound examination^{4,6-8}.

In about 15%-25% of cases, the disease presents as a nodular, nodulo-ulcerative, fungating or polypoid lesions resembling rectal growth or sessile polyp. The colonoscopic finding of our case was a circumferential ulcero-nodular lesion about 5 cm from anal verge. Al Ibrahim et al⁴ in their clinicopathological study of 13 cases showed that solitary ulcer found only in 60% of cases and the other patients presented as multiple ulcers or polypoid lesions. Tendler et al⁵, on the other hand reported that all their 15 patients with SRU presented as polypoid lesions. A study by Tjandler et al⁶ showed that about 45% of their patients presented as polyps and ulcer found only in 29% of patients.

Rarely, colonoscopic findings show only hyperemia, erythematous and thickened rectal mucosa with no evidence of ulcerative or nodular lesion giving pictures similar to that of inflammatory bowel disease. Multiple biopsies from the lesion is a rule in SRU to confirm the diagnosis^{4,6}.

Histopathological examination of rectal biopsy remains the gold standard diagnostic tool for SRU and to exclude other conditions mainly rectal cancer and inflammatory bowel disease. The main histopathological changes that are consistent with SRU include fibromuscular obliteration of the lamina propria, hypertrophy and thickening of muscularis mucosa, glandular crypt distortion, surface serration with evidence of chronic and/or acute inflammation. The presence of inflammatory infiltrates together with non specific symptoms and variable endoscopic findings make the diagnosis and distinguishing SRU from inflammatory bowel disease difficult and challenging^{2,4,7}. In a study by Tjandra et al⁸, they found that 7 patients with actual SRU were misdiagnosed and treated initially as cases of ulcerative colitis.

Several histopathological studies emphasized that fibro-muscular obliteration, abundant deposition of collagen in the mucosa and abnormal extension of smooth muscle fibers with diamond shaped crypts are the main histological features that help differentiating SRU from ulcerative colitis⁹⁻¹².

SRU is very rarely associated with malignancy especially in young patients. The presence of reactive regenerative atypia in the epithelial surface of rectal mucosa, due to ulceration with its associated degeneration and regeneration make some cases of SRU mimic dysplasia and malignancy of the rectum^{13,14}.

It has been found that inflammatory bowel disease mainly ulcerative colitis and rectal malignancy are the most common conditions that mistaken and represent diagnostic dilemma of SRU^{11,14}. Other diagnostic tools such as MRI of pelvis, rectal and endoanal ultrasound, anal manometry, barium enema and defecography are of little help and their results are usually non specific^{4,7,10,13}.

The radiological picture of SRU by MRI characterized by appearance of thickened ulcerated mucosa in the anterior rectal wall which is usually indistinguishable from rectal cancer. However, the presence of rectal prolapsed and location of the ulcer on anterior rectal wall should alert the suspicion of SRU¹⁵. Trans-rectal ultrasound shows accurate measures of the thickness of rectal wall layers and of anal sphincters. Thickened and hypertrophy of muscularis propria indicates chronic mechanical load on the anterior rectal wall which is the main site inflammation and ulceration in SRU. Besides, rectal ultrasound shows absence of relaxation of puborectalis muscle during straining which is one of the suggested pathological mechanism in SRU^{16} . Behera et al¹⁷ found that abnormities of anorectal manometry were more prevalent in patients with SRU.

Ulceration, stricture. granularity, polypoid lesion or normal rectal mucosa may all be recorded on barium enema¹⁸. The role of defecography for patients proved to have SRU is to evaluate the associated defecation disorder as gradual enfolding of the rectal wall toward the lumen to form an intussusceptions or failure of relaxation of pelvic floor musculature during straining as mentioned by Goei et al¹⁹

Treatment of SRU patients is variable from behavioral and ranges and conservative treatment, medical therapy and surgery depending on duration and severity of the disease, education of patients, and presence of rectal prolapse. Modification of lifestyle such as dietary changes including increasing dietary fibers with stool softeners or bulking agents and behavior therapy to stop excessive straining during defecation, regulation of bowel habits and avoiding digital evacuation of the rectum are usually advised for mild cases²⁰. Patients who did not responsd to these conservative measures. biofeedback therapy which is a highly specialized form of behavioral treatment is advised²⁰⁻ ²¹. This mode of treatment entails behavioral modifications such as the suitable posture during defection and reduced the time spent and visits to toilet in addition to avoidance of straining, rectal digitations, and use of laxatives. Attention to any psychological disorders important²¹. Subjective is also improvement in SRU symptoms was



Fig.1: SRU; Hyperaemia of mucosa

noticed in about two third of patients after biofeedback but the improvement was short termed^{22,23}.

Certain medications such as topical steroids, sucralfate, sulfasalazine or mesalazine enemas and recently botulinum toxin has been used with variable results and high recurrence rate^{24,25}.

Surgery is preserved for sever, long standing and refractory cases that failed to improve on behavioral & conservative measures and for patients with rectal prolapse. The surgical options for SRU include mainly excision of redundant sigmoid and rectopexy as what has been done for our patient, or excision of nodular or polypoidal SRU with rectopexy. Proctectomy is usually preserved for patients with severe rectal bleeding and intractable pain when other surgical measures failed to control these symptoms^{7,24}.

CONCLUSION

Histopathological examination of the multiple biopsies taken from different parts of lesion is the corner stone in the definite diagnosis. Treatment of SRU depends mainly on the severity of symptoms and presence of rectal prolapse. The treatment ranges from behavioral and biofeedback therapy for mild to moderate cases. topical medication in form of steroids sulfasalazine, botox injection is the second line of treatment and surgery for severe cases.



Fig. 2: SRU; Solitary ulcer

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Fig. 3: SRU; Multiple ulcers





Fig. 4: SRU; Polypoid & nodular ulcer



Fig 5: MRI Pelvis: SRU with anterior wall rectal prolapse

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