IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 17, Issue 11 Ver. 8 (November. 2018), PP 21-25
www.iosrjournals.org

Solitary Rectal Ulcer Syndrome: Case Report of Two Patients from a Tertiary Care Centre in Manipur, North-East India.

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Abstract: *Solitary rectal ulcer syndrome (SRUS) is an uncommon benign disorder of defecation. The mechanisms of this condition are poorly understood. SRUS forms a component of the spectrum of benign defecation disorders comprising rectal prolapse, proctitis cystica profunda (PCP) and inflammatory polyps. Complications of SRUS include rectal prolapse, intussusception etc. However, malignancy has not been reported following SRUS. The most accepted etiopathogenic mechanism of SRUS is chronic hypoperfusion induced ischemic injury to rectal mucosa. Definitive treatment of SRUS is still unclear varying from conservative management to surgical procedures such as rectopexy or resection and anastomosis. Here we are reporting cases of two young females admitted in the surgical ward of RIMS Imphal presented with complaints of recurrent bleeding per rectum. On examination patients vitals were stable. Pallor was noted. Digital rectal examination and proctoscopy findings were within normal limits. On further evaluation with colonoscopy, solitary rectal ulcer was noted in both patients. Initially ulcer was managed conservatively using stool softeners, bland diet, iron supplements and enema. But symptoms did not subside by conservative methods. Se we planned for surgical intervention in the form of localised anterior resection of rectum and anastomosis using stapler technique. During the post-operative and follow up period patients improved symptomatically. Hence, we are suggesting that localised anterior resection of rectum with primary anastomosis using single or double stapler technique is a good treatment option for SRUS when conservative management fails.*

Key words: *solitary rectal ulcer syndrome, SRUS, rectal prolapse, localised anterior resection*

Date of Submission: 14-11-2018 Date of acceptance: 29-11-2018

I. Introduction

Solitary rectal ulcer syndrome (SRUS) is an uncommon benign disorder of defecation, the mechanism of which is poorly understood. It is characterized by a combination of symptoms, endoscopic findings, and histological abnormalities. It was first described by Cruveihier in 1829, when he reported four unusual cases of rectal ulcers.The term "Solitary ulcers of the rectum" was used by Lloyd-Davis in the late 1930s[1].Most accepted etiopathogenic mechanism of SRUS is chronic mucosalhypo perfusion induced ischemic injury to the rectal mucosa. This is associated with paradoxical contraction of the pelvic floor leading to mucosal prolapse and pressure necrosis of rectal mucosa. Other hypothesis suggests that external anal sphincter produces abnormal pressure gradients in the opposite direction which results in abnormal defecation leading to SRUS[2]. SRUS forms a component of the spectrum of benign defecation disorders comprising rectal prolapse, proctitis cystica profunda (PCP) and inflammatory cloacogenic polyp; the four entities sharing the same clinical and pathological features.

Despite the name, which suggests that it is a specific entity with typical location and presentation, this is not so.Rectal bleeding, constipation, and straining at stool are common presentation, the combination of these symptoms suggesting a local disease in the rectum. Men and women are affected equally, with a small predominance in young women. SRUShas also been described in children and in the geriatric population. The characteristic histological features include surface serration, fibromuscular obliteration, and crypts' distortion. In addition, different vascular changes, such as ectasia, congestion, and hyalinization, can be seen. It is important to note that these features are not pathognomonic of SRUS.This wide spectrum of clinical features, endoscopic findings, and histological features make SRUS a great mimicker of other serious conditions, including adenocarcinoma, inflammatory bowel disease, dysplasia, and adenomatous polyp. Opinion differs regarding the best treatment for this troublesome condition,varying fromconservative management and enema preparations

tomore invasive surgical procedures such as rectopexy, Delorme's procedure, resection and anastomosis. The data on clinical, endoscopic and management spectrum of SRUS is scarce in this region of the world.

We are reporting two cases of solitary rectal ulcer syndrome without proctoptosis who were admitted in the surgical ward of RIMS, Imphal, Manipur.

Case report 1:

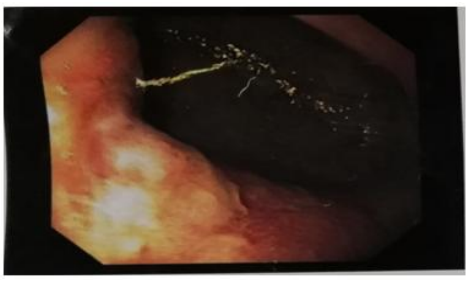
A 15-year-old girl with known case of solitary rectal ulcer presented to OPD with history of constipation (recurrent episodes) and intermittent fresh rectal bleeding with clots for the last 6 months. The bleeding was accompanied by mucus discharge and was associated with tenesmus. This led to a mild decrease in haemoglobinlevel (Hb 9.3). Patient did not give any history of mass per rectum. According to history she was on regular meals which was deficient in fibres and consisted of more proportion of oily foods. There was no history of anal self-digitation, foreign body insertion and there was also no family history of inflammatory bowel disease. Patient was on oral iron preparation, stool bulking agents, enema for constipation and dietary management.

On examination pallor was noted. Vitals were stable at the time of presentation. On abdominal examination, abdomen was soft, non-tender and bowel sounds were heard normally. No organomegaly noted.No ascites as well. Digital rectal examination and proctoscopy findings were non-significant. Biochemical parameters showed Hb 9.3g/dl, liver function test and kidney function tests were normal. Coagulation profile was in normal limits. Ultrasonography of abdomen showeda left sided small sized renal calculus and all other organs appeared normal. Colonoscopyexamination however, showed the presence of an anteriorly located solitary rectal ulcer approximately 10 cm from the anal verge. Biopsy was taken again, and it was consistent with previous finding of solitary rectal ulcer syndrome. In view recurrent episodes of symptoms, failure of conservative management and large size of ulcer,patient was planned for surgery in the form of localised anterior resection with double or single stapler technique.

In view of recurrence of disease following anterior rectopexy procedure, we prefer to go with localised anterior resection. Intra operatively, localised resection of rectum almost 7cm from the anal verge along with6cm proximally from the distal margin done manually. Following resection, primary anastomosis was done using circular stapler. To protect the distal anastomosis, a diversion loop ileostomy was created. Cut section of specimen findings revealed inclusion of all margins of ulcer on the anterior wall of rectum. Post-operative period was uneventful and stoma was functioning well. Oral feeding startedon day 4 of post procedure. Histopathology examination revealed solitary rectal ulcer with adequate resection margins. Patient was discharged and advised to attend in ward after 6 weeks for closure of ileostomy.

Case report 2:

A 24-year-old young lady presented to the emergency department, for the first time, with painless bleeding from the rectum. She had no previous history of such episodes, but she had history of recurrent episodes of constipation for last six months for which she had received treatment with stool softening agents andlaxatives. There was no history of psychiatric illness. On the other hand, the dietary history of the patient was unremarkable.Her general physical examination was unremarkable apart from mild pallor. Digital rectal examination and proctoscopy did not find any abnormalities. Herhaemoglobin concentration was 9 gm/dl.Other biochemical parameters including the coagulation profile were within normal limits. The patient underwent colonoscopy which showed a single ulcerated lesion about 12 cm from the anal verge on the anterior wall of the rectum. Biopsy taken from the ulcerated lesion was suggestive of solitary rectal ulcer. Patient was given a trial of conservative management using stool bulking agents and laxatives for a period of 3 months. In spite of conservative management her symptoms were persisting. So we decided to go for surgical management and she underwent localised anterior resection of rectum with primary anastomosis using single stapler technique. The histopathology specimen of resected specimen was consistent with features of solitary rectal ulcer syndrome.Patient improved symptomatically following the surgery.



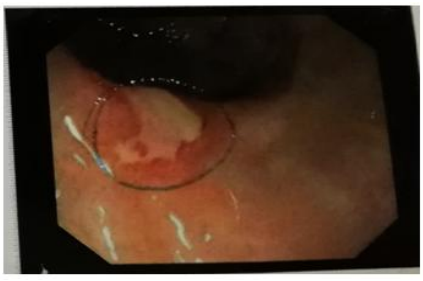


Fig-1 and Fig-2: Colonoscopy images showing solitary rectal ulcer in patient 1 and patient 2

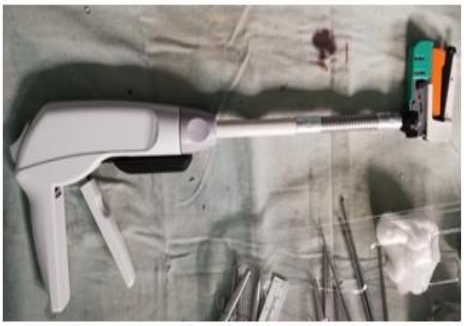


Fig-3: Linear cutter stapler for resection and closure of the distal stump

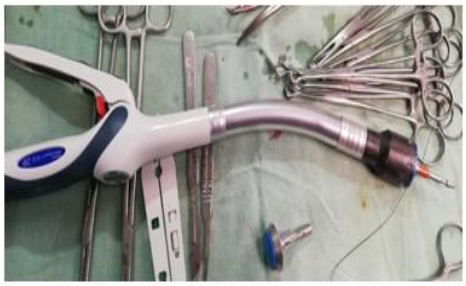


Fig-4: Circular stapler for primary anastomosis of proximal and distal ends of rectum

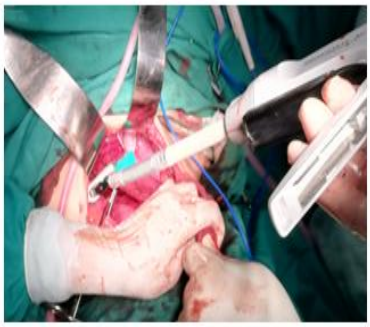


Fig-5: Image showing resection and closure of distal stump

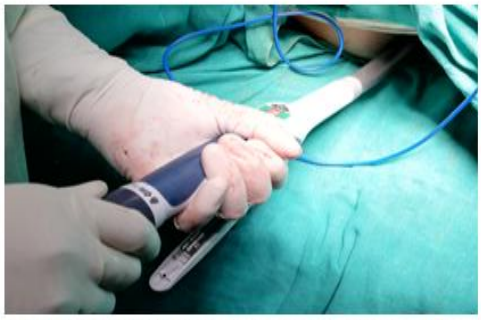


Fig-6: Image showing primary anastomosis using circular stapler

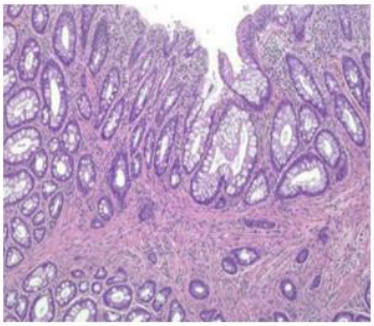


Fig-7: Photomicrograph showing fibromuscular hyperplasia in the lamina propria. (H&E Stain, 10X)

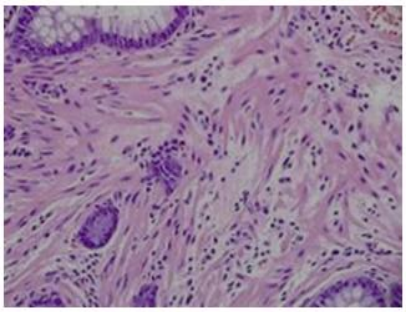


Fig-8: Photomicrograph showing fibromuscular hyperplasia in the lamina propria. (H&E Stain, 40X)

II. Discussion

In this article wenoticed that both patients were females of younger age group which is quite similar to that of previous studies on this subject. Moreover the presenting symptom was recurrent bleeding per rectum, which is the common presentation of solitary rectal ulcer syndrome. The SRUS aetiology, despite many studies and observations, has not been discovered. The most popular hypothesis claims that this is a secondary disease following rectal prolapse (overt or latent, mucosal or complete prolapse) and it is a result of defecation disorders. Disorders of muscle synergy related to defecation are seen in 25–82% of patients with SRUS. Sharara et al[3]estimate that the proportion of defecation disorders in patients is as high as 75%. Another hypothesis related to defecation disorders draws attention to the oversensitivity of rectal mucosa, leading to a continuous feeling of incomplete defecation and excessive rectal tenesmus. The influence of atherosclerotic changes on mucosal ischaemia is also suggested, as well as disorders in cholinergic synapses of the autonomous nervous system, which can be related to diabetic polyneuropathy. It is also suspected that SRUS can be an innate malformation of the hamartoma type. The final SRUS diagnosis is established on the basis of histopathological criteria. A study of samples taken at endoscopy shows changes in all the layers of the rectal wall. The lesions occur about 10 cm away from the anal verge. They are of polymorphic character: ulcerations, polyp-like changes, and flat changes. Polyp changes dominate in a non-symptomatic group, and ulcerations always seem to be symptomatic. Macroscopic changes in SRUS are localised mainly on the rectal anterior wall, and more rarely on the dorsal and lateral wall.

In the above reported cases, SRUS did not coexist with rectal prolapse, which considerably limits treatment options when conservative treatment is ineffective.Lack of uniform agreements about procedures in SRUS also results from non-homogenous and poorly recognised pathogenesis. The therapy initially consists of conservative methods and is usually the end of therapy if there is some improvement or if patients are asymptomatic. Subsequently, it is recommended that patients follow a high-fibre diet to regulate defecation rhythm, perform relaxation exercises of the sphincter muscles, and avoid mechanical procedures leading to local injuries of rectal mucosa. Good results were achieved by using behavioural therapy: training based on biological feedback. This method is based on establishing control of the external rectal sphincter with relation to the patient's biological needs. The aim is to improve defecation technique, to decrease rectal oversensitivity to stimuli, to recover normal mechanism of defecation and thus to make clinical symptoms disappear, to normalise the frequency and quality of defecation, and to heal the lesions in the rectum. Most probably the feedback influences autonomic innervation and as a resultimproves mucous flow and provides favourable conditions for rectal wound healing. Topical treatments, including sucralfate, salicylate, corticosteroids,sulfasalazine, mesalazine and topical fibrin sealant, have been reported to be effective with variousresponses and improvement of symptoms. Sucralfateenema contains aluminium complex salts, which coat the rectal ulcer and form a barrier against irritants, allowing the ulcer to heal. Corticosteroids and sulfasalazine enemas may also help ulcer healing by reducing the inflammatoryresponses. However, these treatments are empirical and have been applied in uncontrolled studies, and their long-term benefits deserve further investigation.

Here the patients were managed conservatively in the form of diet restrictions by avoiding spicy and oily foods, high fibre diets, stool bulking agents and enema to avoid straining at stool there by to improve the healing of ulcer and to prevent exacerbation of condition. Associated anaemia was managed with blood transfusion and iron preparation infusion. The patients were managed conservatively for about 6months with serial sigmoidoscopic evaluation. But the symptoms were persisting even after the conservative approach. Rectal ulcers were comparatively larger in size and due to the unavailability non-operative management like photocoagulation or thermal ablation using argon photocoagulation we planned for surgical intervention in the form of localised resection and anastomosis[4,5]. Due to higher up position of ulcer location,it supports our treatment option rather than anterior rectopexy.Surgery remains an option for patients who are not responsive to conservative measures and biofeedback. Surgery is warranted in almost one-third of adults with associated rectal prolapse; in children this has only been described in case reports. Surgical treatments include excision of the ulcer, treatment of internal or overt rectal prolapse, and de-functioning colostomy.

The indication for surgery is failure of conservative treatment to control severe symptoms, and the aim is to avoid formation of colostomy as a primary operation. Sclerotherapy injection into the submucosa or retro-rectal space with 5% phenol, 30% hypertonic saline or 25% glucose and perianal cerclage is effective in treating rectal prolapse. A therapeutic role of botulinum toxin injection into the external anal sphincter for the treatment of SRUS, and constipation associated with dyssynergia of defecation dynamics has also been reported by Keshtgaret al[6-10]. The effect of botulinum toxin lasts approximately for 3 months. In children, laparoscopic rectopexy using a polypropylene mesh on each side of the rectum, fixed to sacral promontory with a nonabsorbable suture, has been used successfully to treat SRUS. Furthermore, for full-thickness prolapse, mucosal resection (Delorme's procedure) or perineal proctectomy (Altemeier's procedure) has been advocated. Local excision of polypoid rectal ulcer and rectopexy for overt rectal prolapse, howeverhave a higher longterm cure rate. Proctectomy may be required in patients with intractable rectal pain and bleeding, who have not responded to other surgical treatments[11]. Based on postoperative evacuation defecography studies, it has been shown that rectopexy alters rectal configuration and successfully treats rectal prolapse in SRUS, and that a prolonged preoperative evacuation time is predictive of poor symptomatic outcome. When the above measures fail, mucosal-sleeve resection with coloanal pull-through or a diverting colostomy should be considered. The evidence regarding which approach is first-line for SRUS is unclear. However localised resection seems popular with a success rate of 42%-100%.

Localised anterior resection of rectum with primary anastomosis using single stapler or double stapler technique was adopted in these patients[12-16]. Intraoperative and post-operative period were uneventful in these patients. One patient with diversion loop ileostomy underwent closure after 4 weeks. Both the patients are doing well.

III. Conclusion

The exact treatment option of solitary rectal ulcer syndrome remains unclear. Here we are suggesting,localised anterior resection of rectum with primary anastomosis using single stapler or double stapler technique is a good treatment option for SRUS when conservative measures fail.

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Dr. Sreejith V "Solitary Rectal Ulcer Syndrome:Case Report Of Two Patients From A Tertiary Care Centre In Manipur,North-East India. ""IOSR Journal of Dental and Medical Sciences (IOSR-JDMS), vol. 17, no. 11, 2018, pp 21-25.