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Case Report
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A rare cause of chronic rectal bleeding in children; solitary rectal ulcer: case report

Çocuklarda kronik rektal kanamanın nadir bir sebebi; Soliter rektal ülser: Olgu sunumu

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Solitary rectal ulcer causing lower gastrointestinal bleeding is extremely rare in children. Rare presentation, non-specific symptoms, insufficient experience, and characteristics mimicking other rectal diseases may cause misdiagnosis or delay of diagnosis in some pediatric patients. Here, we report a 10-year-old boy with solitary rectal ulcer diagnosed two years after onset of the symptoms who responded well to the conservative therapy, including high-fiber diet, laxatives, defecation training, and sucralfate enema.

Key Words: Children; gastrointestinal bleeding; rectal; solitary; ulcer.

Alt gastrointestinal kanamaya neden olan soliter rektal ülser, çocuklarda oldukça nadir görülür. Nadir görülmesi, özgün olmayan bulgularla ortaya çıkması, bu konuda yeterli deneyimin olmaması ve çeşitli rektal hastalıkları taklit etmesi, bazı çocuk hastalarda yanlış veya gecikmiş tanıya neden olabilmektedir. Burada, belirtilerin başlangıcından 2 yıl sonra tanısı konan, yüksek lifli diyet, laksatif, tuvalet eğitimi ve sükralfat lavman tedavisine iyi yanıt veren soliter rektal ülserli 10 yaşındaki bir olgu sunuldu.

Anahtar Sözcükler: Çocuk; gastrointestinal kanama; soliter; rektal; ülser.

Lower gastrointestinal (GI) bleeding is a common problem in children, and it resolves spontaneously in many instances.[1,2] Although solitary rectal ulcer syndrome (SRUS) causing lower GI bleeding is relatively well documented in adult series, pediatric cases are extremely rare, and only a few case reports have been published. The incidence of SRUS was reported by El-Khayat et al. and by Mandhan to be 1.5% in children with rectal bleeding and 3.5% within chronic lower GI bleeding, respectively.[1,3] However, the exact incidence is unknown in childhood.

Rare presentation, non-specific symptoms, insufficient clinic and pathologic experience, and characteristics mimicking other rectal diseases are the causes of failure and delay of diagnosis in some pediatric patients with SRUS.[4,5] Tjandra and Rao et al. reported the mean period between the onset of the symptoms and correct diagnosis to be 7.8 years and 4.5 years, respectively.[5,6]

We report a 10-year-old boy with SRUS diagnosed two years after the onset of the symptoms and we discuss his clinical course in light of the literature.

CASE REPORT

A 13-year-old boy with a two-year history of rectal bleeding, mucous defecation and chronic constipation was admitted. He reported straining defecation, and no rectal digitation or rectal prolapsus with detailed history. No pathologic finding was detected on rectal examination. Plain abdominal X-ray, contrast barium enema and investigation for parasitic infestation revealed no pathology. The rectoscopic and colonoscop-

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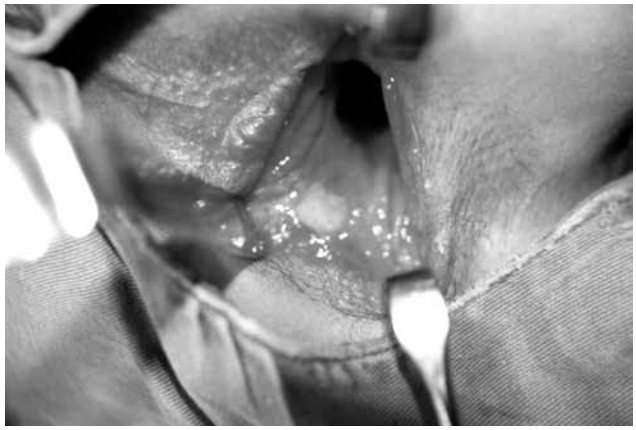

Fig. 1. The rectal examination shows a 2 x 3 cm ulcer covered with fibrinous exudates on the posterior rectal wall, 2 cm proximal to the dentate line.

ic examination showed a 2 x 3 cm ulcer covered with fibrinous exudates on the posterior rectal wall (Fig. 1) and two ulcers of 1 x 2 cm and 2 x 4 cm on the anterolateral rectal wall 2 cm and 10 cm proximal to the dentate line, respectively, as well as 2 x 4 cm hyperemia located 25 cm above the dentate line. The most distal lesion was excised with normal mucosa surrounding the ulcer. Histopathological examination revealed wide superficial mucosal ulceration, mixed inflammatory cell infiltration, vascular proliferation, increased fibrous tissue, and extended glandular structures on the lamina propria indicating a SRUS (Fig. 2). The patient was treated conservatively with a high-fiber diet, laxative, defecation training, and 10% solution of sucralfate enema twice daily for two months. A second rectosigmoidoscopy was performed after six months, and no pathologic lesion could be detected. He became completely asymptomatic during the three-year follow-up.

DISCUSSION

Solitary rectal ulcer syndrome (SRUS) was first described by Cruveilhier in 1830.[3,7,8] Its etiology is still unclear.[4,9-11] Previously, rectal mucosal ischemia and trauma were reported to be related in the etiology of SRUS.[7,11-13] Defecation straining causes puborectalis relaxation allowing the stool passage in the normal situation. Rutter demonstrated increased puborectal electromyographic activity in patients with SRUS and cited this abnormality for the pathogenesis of SRUS with rectal prolapsus.[14] Womack et al.[15] reported in an adult study occult or manifest rectal prolapsus in 94% of patients with increased intrarectal pressure and electromyographic activity of the external anal sphincter. They considered that the increased activity of the external anal sphincter requires high intrarectal pressure to perform defecation. Thus, they claimed that rectal prolapsus with high intrarectal pressure might be the cause of the mucosal damage. Satish et al.[6] found dyssynergia, rectal hypersensitivity, paradoxical anal contraction, and impaired evacuation in 82% of their patients. Defecography may show the evident and occult rectal prolapsus, delayed evacuation of radiocontrast, and enterocele.[6,7,13] For technical reasons, we did not perform a defecography or anorectal

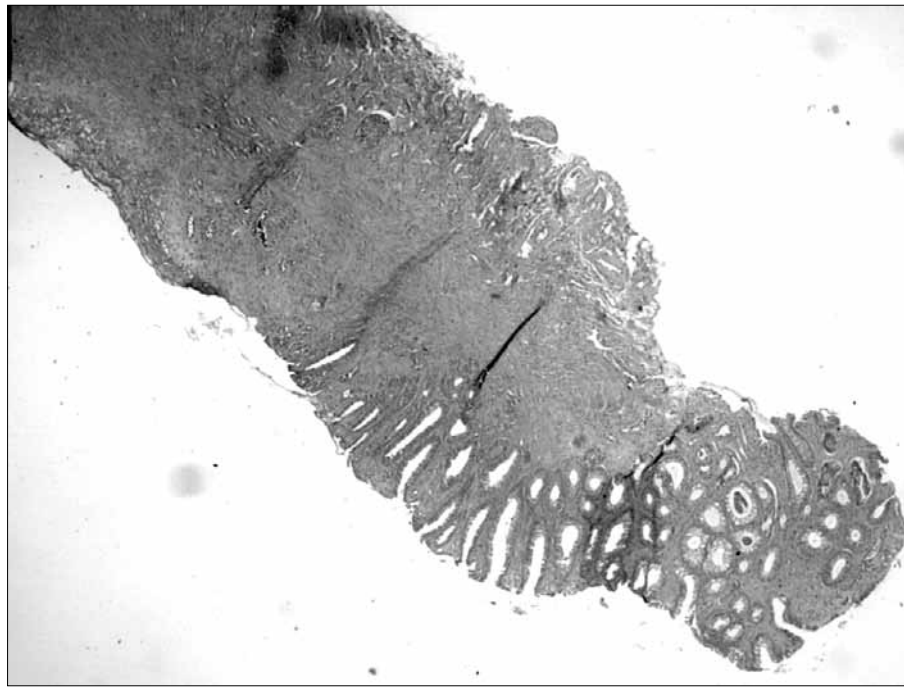

Fig. 2. Widespread superficial epithelial ulceration, mixed inflammatory cell infiltration with vascular proliferation and increased fibroblastic activity are seen in the histopathological evaluation.

manometric study in our patient; there was a history of straining, but no evidence of rectal prolapsus. Some trials suggested digital trauma to the rectal mucosa as a possible factor in developing SRUS.[6,11-13,16,17] Based on these reports, high intrarectal pressure with overactive external sphincter, abnormal contraction of the puborectalis muscles, and anal digitations lead to venous congestion, mucosal ischemia and ulceration.[12,13,16-19] However, none of the affirmed hypotheses can exactly explain the pathogenesis.[4,9,11]

Although SRUS presents generally with rectal bleeding, mucus passage, rectal pain, and tenesmus, additional complaints such as constipation or diarrhea, prolonged straining, altered bowel habits, and rectal prolapsus may be discovered once a detailed history is obtained.[5,7,8,10,18] Gabra et al.[20] reported a case of SRUS who presented with severe rectal stricture. In 26% and 21% of a series of patients with SRUS including children and adults, Tjandra et al.[5] reported presence of psychiatric disorders and absence of symptoms, respectively. The primary symptom in our case was rectal bleeding, which is also reported to be most common symptom in the pediatric age group.

There is some confusion about the term SRUS. The lesions may not be only solitary but can also be multiple or circumferential. Multiple ulcers have been reported in 30% of patients.[17] The endoscopic appearance of SRUS can be of three macroscopic types: ulcerative, polypoid and hyperemic. The ulcerative form is the most common in children (60%). The site of the lesions does not differ between the three types. The most frequent sites are the anterior or anterolateral wall of the rectum, 5 to 10 cm proximal to the dentate line. Ulcers are usually 1 cm to 1.5 cm and are encircled with hyperemic mucosa.[8,12,16-19] Contrary to this general information, three of the four ulcers in our patient were larger than expected and their localization was outside the 5 to 10 cm area above the dentate line, which is the traditional localization.

Ulcerative and hyperemic types of SRUS can be confused with inflammatory bowel diseases, especially with Crohn's disease.[4,5,7,16,17,19,20] In previous reports, the macroscopic appearance of the hyperplastic type could not be distinguished from inflammatory polyp of the rectum or villous adenoma.[4,7,19] The characteristic histopathological findings of SRUS that provide differentiation from other diseases are obliteration with increased fibroblastic activity and presence of collagen deposition in the lamina propria, distortion of crypt architecture, lack of epithelial dysplasia, regenerative changes in crypt epithelium, and muscle fibers derived from muscularis mucosa in the surrounding tissue.[1,7,9,12,13,16,17]

The diagnosis of SRUS is established by appraising symptoms together with endoscopic appearance and histopathologic findings. Since it is quite rare in children and symptoms are not characteristic, clinicians, endoscopists and pathologists should keep SRUS in mind to prevent delayed diagnosis. Therefore, clinicians should inquire at length about the bowel habits in children with rectal bleeding because defecation habits change towards constipation, which was approved as one of the etiologic factors of SRUS in school-aged children with previously regular habits. Thus, constipation history can be helpful in reaching the correct diagnosis.

There is no agreement on the treatment of SRUS.[7,9,12,21] Whatever the treatment method of choice, the goal of the treatment is to recover bowel habits and improve rectal ulcers. The initial stage of the treatment strategy must be a conservative approach, which includes defecation training for the patient and behavioral modification unless there is evidence of full-thickness or mucosal rectal prolapsus that requires surgical intervention. Defecation training including avoidance of straining and anal digitations, biofeedback, high- fiber diet, and some medication are principal components in the conservative treatment.[4,7,8,12,13] Some authors have proposed topical steroids, sulfasalazine or local excision of the polypoid-type lesion in patients with SRUS; however, there are studies reporting these treatments as being insufficient.[5,7-9] Zargar et al.[21] obtained clear improvement in clinical symptoms and the endoscopic appearance with sucralfate enemas. Nevertheless, no marked histological change was found. Many clinicians still use sucralfate retention enemas for the treatment of SRUS, with limited success.[21,22] We obtained complete recovery in both clinical complaints and endoscopic picture with conservative treatment including high-fiber diet, laxatives, defecation training, and sucralfate in our patient two months after onset of the treatment.

Rao[6] demonstrated that biofeedback therapy improves dyssynergia, bowel symptoms, and mucosal changes and reforms defecation dynamics. They suggested the biofeedback therapy for routine management of patients with SRUS, especially in those who are refractory to the medical treatment.

For the patients with accompanying rectal prolapsus or those who do not respond to the medical treatment, many surgical techniques such as Ekehorn's and Delorme's sacral rectopexy, the Ripstein procedure and low anterior resection were described previously.[4,8,5,19] Bonnard et al.[18] performed laparoscopic rectopexy in children with SRUS who did not improve with medical treatment. However, there is still little knowledge about results of surgical treatment in children.

In conclusion, SRUS must be kept in mind in children presenting with chronic rectal bleeding who are refractory to treatment. Endoscopic examination is

necessary to prevent delayed diagnosis or misdiagnosis. Increase in the number of reported patients who are diagnosed and treated successfully will expand our knowledge on the diagnosis and treatment of SRUS in pediatric cases.

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