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Solitary rectal ulcer syndrome in a child: An unusual cause of rectal bleeding

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Summary

Solitary rectal ulcer syndrome is an uncommon cause of rectal bleeding in children. In this report a 10 years old child presenting with rectal bleeding due to this syndrome will be presented and the etiology, diagnosis and treatment will be reviewed.

Key words: Solitary rectal ulcer syndrome, rectal bleeding.

Introduction

Rectal bleeding is a common symptom in pediatric surgical pratice. The causes of rectal bleeding are well-known in different pediatric age groups. In children older than 1 year of age; polyps, Meckel's diverticulum, inflammatory bowel disease, trauma, rectal prolapse, infectious diseases may be responsible of rectol bleeding. Solitary rectal ulcer syndrome is a very rare cause of rectal bleeding in children, even in adults. Since 10% of the the cases with rectal bleeding are with unknown etiology, diagnosis and treatment of this condition still remains a challenge [8]. In this report an unusual cause of rectal bleeding in a child due to solitary rectal ulcer syndrome will be presented and the literature reviewed.

Case Report

MC. a ten-year-old boy was admitted to Şişli Childrens' Hospital, Pediatric Surgical Clinic with chief complaints of passage of fresh blood mixed with stool and perineal pain. He presented with a one year history of intermittent rectal bleeding which recently had increased in frequency. Despite the child's discomfort, the parents had been reluctand in seeking for medical help.

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Rectal examination done by the resident on the outpatient clinic was noted as normal. Routine laboratory tests were within normal limits. The patient was hospitalized for rectoscopic examination according to the clinic's rectal bleeding protocol. Rectoscopy confirmed an ulcerated, hyperemic lesion with well demarcated edges 2 cms. in diameter, on the anterior wall of the rectum, 6 cms. from the dentate line. The surrounding mucosa was normal and a bleeding site could not be determined. Rectoscopy upto 11 cms. demonstrated no other abnormality. A wide local excision was attemmpted and during the procedure profuse bleeding occured. The mucosal defect was repaired with 3/0 silk sutures. Histology revealed fibrosis of the lamina propria, ulceration and hypertrohy of the muscularis mucosa and distorted crypts, characteristis of "solitary rectal ulcer syndrome". No rectal bleeding occured on the postoperative period and the patient was discharged with laxatives. Rectal examination ten days later revealed a normal rectal mucosa with no recurrent ulceration. The patient remains symptomless and receives no therapy.

Discussion

Solitary rectal ulcer syndrome is a rare cause of rectal bleeding in children. This condition occurs more frequently in young adults with an equal sex incidence.

The first description of benign rectal ulceration was by Cruveilhier in 1983 [1]. The term "solitary rectal ulcer" was introduced by Lloyd-Davies in 1973 [3]. A detailed description of the disease was by Madigan and Morson in 1969 in which they reviewed 68 cases collected at St. Mark's Hospital [4].

Patients with this condition mainly present with rectal bleeding. Other presenting symptoms may be passage of mucus on defecation, perineal pain, inceraesd frequency of defecation and tenesmus. These symptoms may last for many years and become a disturbing problem for the patient [2,3,4,5,6].

The etiology of this syndrome is still controversial. Rutter in 1975 demonstrated electromyograhpically a failure in the ralaxation of the puborectalis muscle in those who strain during defecation, leading to anterior rectal mucosal prolapse into the anal canal where it becomes traumatized and ulcerated [6]. Schweiger and Alexander-Wililams demonstrated proctoscopically that occult rectal prolapse was present in a group of their patients suggesting an association of rectal mucosal prolapse with solitary rectal ulcer syndrome [7]. We could not obtain a history of straining and rectal prolapse in this case and the patient mainly presented symptoms of rectal bleeding and perineal pain which may be present with any other anorectal disease.

Rectoscopy characteristically demonstrated an ulcer with clear demarcation from the surrounding mucosa located on the anterior rectal wall. In some cases it may simulate proctitis with a granular, inflammed rectal lesion [2,6]. The characteristic histologic appearance of the lesion described by Madigan and Morson is:

1. Fibromuscular obliteration of lamina propria
2. Hypertrophy of muscularis mucosa
3. Misplacement of glands deep to muscular is mucosa [2,4].

All of these criteria were present in this case which led to diagnosis. The differential diagnosis includes, adenocarcinoma of the rectum, Crohn's disease, ulcerative colitis and villous adenoma [2,6,9].

No satisfactory treatment is available for this chronic disorder. Following pathologic diagnosis these patients must receive medical therapy aimed to reduce irritation of the rectal mucosa in first place. Program of the conservative therapy mainly consists of laxatives. Those who fail to respond to conservative therapy are considered for surgery. Colostomy, wide local excision and abdominal rectopexy has been performed in adult patients with this syndrome. While recurrence following colostomy and wide local excision has been high, results of abdominal rectopexy have been promising particulary in adult patients with overt rectal prolapse [2,7,9]. On the other hand conservative theraphy should be pereferred to surgical therapy in the pediatric age group. In this patient a wide local excision was attempted, because of a malignanacy suspection. However the patient has responded well to this surgical procedure and still remains symptomless for two months.

Conclusion

Solitary rectal ulcer is a rare cause of rectal bleeding. Since 10% of the cases with rectal bleeding are still of unknown etiology and the number of cases published are incerasing in number every day, this condition must be considered carefully during examination. For these reasons to avoid misdiagnosis, manual rectal examination and rectoscopy of patients with rectal bleeding must be performed by experienced hands.

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