

## Observational Study

## Solitary rectal ulcer syndrome: Is it really a rare condition in children?

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### Abstract

**AIM:** To evaluate the clinicopathologic characteristics of the children with solitary rectal ulcer.

**METHODS:** Fifty-five children with a confirmed diagnosis of solitary rectal ulcer were studied in a period of 11 years from March 2003 to March 2014. All data were collected from the patients, their parents and medical records in the hospital.

**RESULTS:** From 55 studied patients, 41 were male (74.5%) and 14 female (25.5%). The mean age of the patients was  $10.4 \pm 3.7$  years and the average time period from the beginning of symptoms to diagnosis of solitary rectal ulcer was  $15.5 \pm 11.2$  mo. The most common clinical symptoms in our patients were rectal bleeding ( $n = 54$ , 98.2%) and straining during defecation or forceful defecation ( $n = 50$ , 90.9%). Other symptoms were as follows respectively: Sense of incomplete evacuation ( $n = 34$ , 61.8%), mucorrhea ( $n = 29$ , 52.7%), constipation ( $n = 14$ , 25.4%), tenesmus and cramping ( $n = 10$ , 18.2%), diarrhea ( $n = 9$ , 16.4%), and rectal pain ( $n = 5$ , 9.1%). The colonoscopic examination revealed 67.3% ulcer, 12.7% polypoid lesions, 10.9% erythema, 7.3% both polypoid lesions and ulcer, and 1.8% normal. Most of the lesions were in the rectosigmoid area at a distance of 4-6 cm from the anal margin. Finally, 69.8% of the patients recovered successfully with conservative, medical and surgical management.

**CONCLUSION:** The study revealed that solitary rectal ulcer is not so uncommon despite what was seen in previous studies. As the most common symptom was rectal bleeding, clinicians and pathologists should be familiar with this disorder and common symptoms in order to prevent its complications with early diagnosis.

**Key words:** Rectal bleeding; Children; Solitary ulcer; Colonoscopy; Forceful defecation

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**Core tip:** What is known? (1) solitary rectal ulcer is considered a rare condition in children; (2) there is a small number of case report and small case series in pediatric age group in the literature; and (3) this disorder has not been well known in children, so, their symptoms can be confused with other more common diseases. What is new? (1) this study reveals that solitary rectal ulcer is not so uncommon in children; (2) to the best of our knowledge it is the largest pediatric series in the world; and (3) high index of suspicion is needed to think about and diagnosis of this disorder.

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## INTRODUCTION

Solitary rectal ulcer has been defined as an infrequent but benign disorder of rectal and sigmoid region which is diagnosed on the basis of clinical symptom and histologic findings<sup>[1,2]</sup>. Although solitary rectal ulcer is a relatively common disorder in adults, it has been reported as a rare disease in children, so it can be misdiagnosed and troublesome in pediatric cases<sup>[3]</sup>.

The exact cause is not known, but several causes such as trauma, rectal prolapse, ischemia, behavioral disorders such as excessive straining during defecation and rectal manipulation, sexual abuse and disharmony of the pelvic floor muscles during defecation may be involved. Patients with this disorder exhibit a range of symptoms; however, a small number of patients can be asymptomatic<sup>[2,4-6]</sup>.

The clinical symptoms in children are similar to adults, but since this disorder has not been well known in children, their symptoms can be confused with more common diseases. For instance, the obstructive symptoms can be viewed by parents as constipation, rectal bleeding may be related to the anal fissure or causes such as juvenile polyps or tenesmus and also rectal bleeding, may be incorrectly diagnosed as inflammatory bowel disease<sup>[3]</sup>.

Colonoscopic findings are not specific and can be

similar to other disorders of the rectal and anal area<sup>[2]</sup>. The histological study of solitary rectal ulcer has a distinct diagnostic appearance including thickening of the mucosal layer with disrupting crypts structure, infiltration of the lamina propria with fibroblasts, muscle and collagen fibers that lead to hypertrophied and disrupted muscularis mucosa which look like fibromuscular obliteration. Diagnosis of solitary rectal ulcer is usually based on the clinical history and histopathological changes from the rectal biopsy.

Before treatment, it is very important to differentiate solitary rectal ulcer from other disorders of the rectum including cancer, Crohn's disease, granulomatous diseases like lymphogranuloma venereum using laboratory facilities<sup>[7]</sup>.

Many treatments have been tried so far, but there is no strong evidence for their effects<sup>[7]</sup>. It seems that the medical and surgical treatments are not very effective due to the high incidence of recurrence<sup>[8]</sup>.

The first step of treatment is conservative therapy including dietary fiber, fluid intake, behavioral adaptation and use of laxative, which has not been proven to have long term benefits<sup>[2,5]</sup>.

Enema with sucralfate can cover the wounds and act as a protection against harmful substances to heal the wound. Injection of sclerosing agents into the submucosa or rectal space has been useful for the treatment of rectal prolapse in some cases, but the benefits of long term uses need further investigation<sup>[5]</sup>.

Most patients with solitary rectal ulcer have a satisfactory prognosis with treatments such as behavior change in their childhood. Follow up of these patients is necessary in order to improve and sustain treatment method to prevent morbidity and recurrences which lead to the disease progresses to adulthood<sup>[3]</sup>. The aim of this study was to assess the children diagnosed with solitary rectal ulcer in Southern Iran.

## MATERIALS AND METHODS

All of the children including 55 (14 females and 41 males) with final diagnosis of solitary rectal ulcer in Pediatric Gastroenterology Section of Nemazee Hospital affiliated to Shiraz University of Medical Sciences from March 2003 to March 2014 were studied.

The diagnosis of solitary rectal ulcer in our patients was based on the clinical history, colonoscopic and histopathological findings of rectosigmoid biopsies.

The colonoscopies were done under general anesthesia and biopsies were taken from normal and abnormal looking mucosa and all lesions of the rectum and sigmoid colon in all patients. Histopathologic evaluation of all biopsy samples was done by a gastrointestinal pathologist (BG).

After confirming the diagnosis, their information including age and sex, clinical presenting symptoms, colonoscopic and histopathologic findings and also therapeutic modalities and responses to their treatment were retrieved from their registered medical records

and also by phone calls. All of the information obtained from patients' records was registered into the designed collecting data forms; then the analysis of data, was performed according to their age and sex, presenting symptoms, colonoscopic and histopathologic findings and treatment outcome.

This study was approved by the Research Ethics Committee of Shiraz University of Medical Sciences and informed consent was obtained from all the parents or legal guardians.

## RESULTS

In these 11 years, 55 children with the final diagnosis of solitary rectal ulcer were evaluated. The mean age of the onset of symptoms in the patients was  $9.2 \pm 3.4$  years. The mean age of the patients at diagnosis was  $10.4 \pm 3.7$  years. The average time from the onset of symptoms to final diagnosis was  $15.5 \pm 11.2$  mo. The youngest and oldest patients at the onset of symptoms were 1.5 and 17 years, respectively. From 55 patients, 14 were female (25.5%) and 41 male (74.5%).

The first clinical finding in all patients except one was rectal bleeding. One patient presented with a history of passing of mucoid stool and had no history of rectal bleeding during the illness.

The most common presenting clinical symptoms of the patients was rectal bleeding ( $n = 54$ ; 98.2%) and then straining during defecation or forceful defecation ( $n = 50$ ; 90.9%). Sense of incomplete evacuation was reported in 34 patients (61.8%) and 29 cases had a history of passing mucoid stool (52.7%). Change of bowel habits was seen in some patients as 14 patients were suffering from constipation (25.4%) and 9 patients had a history of diarrhea (16.4%). Although abdominal pain is not an obvious symptom of the disease, 18.2% of the patients were suffering from this problem. Three patients (5.4%) had a history of anal manipulation with fingers, 1 had a history of sexual abuse, and 4 patients (7.3%) had a history of rectal prolapse and the sensation of a mass during defecation.

The colonoscopic studies of 55 patients as recorded in their charts were examined. Colonoscopy was normal in 1 of the patients and showed no specific findings (1.8%). Colonoscopy of 37 patients showed ulcers (67.3%). Among the ulcers, 3 were superficial (8.1%), 1 was circumferential (2.7%), 1 was reported as linear (2.7%), and 1 of them was clean based (2.7%). The others (83.8%) were white based ulcers with erythematous borders and exudates. Seven patients had polypoid lesions (12.7%) which were pedunculated in 3 patients (42.8%). Four patients had both ulcers and polypoid lesions (7.3%). Colonoscopy of 6 patients only showed erythema (10.9%) and there was no obvious ulcer. In colonoscopy of 37 patients only one lesion was seen (67.3%), 7 patients had two lesions (12.7%) and 10 patients showed more than two lesions (18.2%).

The ulcers were registered with different sizes. Most of them were 5-15 mm in diameter.

**Table 1** Different treatment protocol and response rate in children with solitary rectal ulcer

| Treatment protocol           | Number | Percent |
|------------------------------|--------|---------|
| Conservative treatment       | 11     | 21.6%   |
| Asacol suppositories         | 23     | 45.1%   |
| Sucralfate enema             | 9      | 17.6%   |
| Alcohol injection            | 2      | 3.9%    |
| Methylprednisolone injection | 1      | 1.9%    |
| Rectopexy                    | 5      | 9.8%    |

All of the ulcers were localized in the rectosigmoid region. The distance of most of them from the anal margin was 4 to 6 cm, but it was about 13 cm in one of the patients and 20 cm in another one.

The histology findings of patients included ulcer, granulation tissue, inflammation, increasing collagen bands and fibrosis in lamina propria, and sometimes elongation of the crypts, fibrosis, reduction in goblet cells and hyperplastic changes.

The therapeutic information of 51 patients was provided from their reports and by phone calls. Different treatments were offered for different patients based on the severity of the symptoms. Conservative measures including avoidance from excessive straining during defecation, use of high fiber diets and fluids, and use of laxatives (if they had constipation) were recommended to all patients. Eleven patients (21.6%) became completely symptom free with only recommended conservative treatments. In 23 patients (45.1%) who had not responded to the conservative treatments, the use of Asacol suppository was recommended. Nine patients (17.6%) were treated with Sucralfate enema.

The alcohol injection was carried out for 2 patients (3.9%) who had rectal prolapse. Methylprednisolone was injected around the lesion in 1 of the patients. Five patients (9.8%) with persistent solitary rectal ulcer and rectal prolapse were finally treated with laparoscopic ventral rectopexy (Table 1).

From 55 patients, 43 were followed by medical reports and also with phone calls. Thirty patients (69.8%) showed a significant improvement during the follow-up and their symptoms disappeared. Most of these patients (28 patients) had responded to the conservative treatments, Asacol suppositories, and Sucralfate enema and 2 patients were treated with rectopexy. Thirteen patients had still their symptoms (30.2%) at the time of the study.

## DISCUSSION

Solitary rectal ulcer is an unusual and uncommon disorder of rectosigmoid region which is mostly seen and reported in adults and is less common in children. This disorder is diagnosed with clinical findings, colonoscopy findings and histology changes<sup>[2]</sup>.

In this study, 55 children with the final diagnosis of solitary rectal ulcer were evaluated in 11 years, and to the best of our knowledge it is the largest pediatric

series in the world.

Most of the patients (74.5%) were male in this study which is similar to other studies and reports on the children with solitary rectal ulcer. In the study by Perito *et al*<sup>[9]</sup>, 9 out of 15 patients were male. Five of 6 children were male in Urganci *et al*<sup>[8]</sup> study and 9 out of 12 patients were also male in Dehghani *et al*<sup>[10]</sup> study, but in Blackburn *et al*<sup>[3]</sup> study, from 8 affected children, 4 were female and 4 were male. However, this predominance in males is only in the pediatric age group and the prevalence of disorder between male and female is the same in adults. It has been even mentioned in some articles that it has more prevalence in the women; so that, from 68 studied patients in Madigan and Morson<sup>[7]</sup>'s study, 33 were male and 35 were female.

In this study, the youngest and oldest patients were 1.5 and 17 years at the onset of the symptoms. The mean  $\pm$  SD of the patients was  $10.4 \pm 3.7$  years and the average of time from the beginning of symptoms to diagnosis of solitary rectal ulcer was  $15.5 \pm 11.2$  mo, these results are similar to Blackburn *et al*<sup>[3]</sup> study in which the mean age of children was 9.87 years and the average time from the onset of the symptoms to diagnosis was 1.73 years. In Urganci *et al*<sup>[8]</sup> study, the average of time from the onset of symptoms to the diagnosis of disease was reported as 4.7 years; this is longer than this study. In Suresh *et al*<sup>[11]</sup> study, the age of the youngest patient studied was 1.5 years which is similar to this study.

In this research, the first presenting symptom in most patients was rectal bleeding (98.2%) and then excessive straining during defecation (90.9%). Other problems in order of their prevalence included the sense of incomplete evacuations, mucoid stools, constipation, abdominal pain and tenesmus, diarrhea, and rectal pain. In the study by Suresh *et al*<sup>[11]</sup> the most common clinical finding of the patients have also been reported as rectal bleeding which has been seen in all 22 patients, but it is expressed that need for blood transfusions in this disorder is low. In another study by Madigan and Morson, the most common clinical symptoms were bleeding from the anus (91%), then mucorrhea, rectal pain, diarrhea and lower abdominal pain<sup>[7]</sup>. In another study by Blackburn *et al*<sup>[3]</sup> all of the patients had the history of straining on defecation and 7 of 8 patients had rectal bleeding history. Other symptoms in order of frequency were sense of incomplete evacuation, tenesmus, mucus excretion, constipation, diarrhea and manipulation of the anus for defecation, respectively<sup>[3]</sup>. According to this study and other related studies, it seems that rectal bleeding is the most common symptom; other less common symptoms were mucorrhea and straining during defecation which can be easily obtained from the patient's history.

The main causes of this disorder are still unknown. In previous studies, it has been concluded that several factors lead to this disorder. In most of the previous studies, it has been stated that the main mechanism

of this disorder is mostly excessive straining during defecation which leads to increased intra-abdominal pressure which in turn causes protrusion of the anterior wall of the rectum into the anal canal and puborectalis muscle contraction and continuation of this state leads to trapping the mucus membrane of the anterior wall of the rectum, edema and hyperemia and finally hypoperfusion, ischemia and ulceration<sup>[12]</sup>. In the present study, more than 90% of patients had excessive straining during defecation which can confirm this problem. In addition to these, 3 patients had the history of anal manipulation with fingers, 1 had the history of sexual abuse and 4 patients had the history of rectal prolapse and sensation of a mass during defecation which is considered as causes of this disorder due to mucosal trauma.

De la Rubia *et al*<sup>[1]</sup> have reported ischemia as the etiology of this disorder because of the lack of trauma in the studied patients and histologic changes in the course of the disease and have suggested that continuous contraction of the puborectalis muscle during defecation can cause hyperemia, edema, necrosis and ulcer in the mucous membrane of the rectum. Womack *et al*<sup>[13]</sup> also concluded that the combination of rectal prolapsed and high pressure during defecation cause instability between intra-abdominal pressure and into the rectum which leads to ruptured submucosal vessels and mucosal necrosis<sup>[13]</sup>. In Dehghani *et al*<sup>[10]</sup> study, traditional way of defecation has been proposed as the cause of solitary rectal ulcer, which leads to protrusion of the anterior wall of the rectum into the anal canal and then hyperemia, edema and ulcer.

Therefore, according to these studies and the results of the present study, it can be concluded that the combination of high rectal pressure during defecation, the hidden prolapse and insufficient contraction of the puborectalis muscle in addition to trauma which cause direct damages, leads to hyperemia, ischemia, and finally ulceration of the rectal wall and the traditional way of defecation in our geographic region in Iran, can intensify these factors; it is the reason for high prevalence of this disease. It has been recommended that defecography and anorectal manometry should be performed in all children with solitary rectal ulcer to define the primary pathophysiological abnormality and to select the most appropriate treatment protocol. These evaluations were not performed in this work.

In the present study, the most common findings in the colonoscopic examination of the patients were ulcer (67.3%), polypoid lesions (12.7%), and mucosal redness and erythema (10.9%). Four patients had both ulcer and polypoid lesions (7.3%) and 1 colonoscopy failed to show any specific finding (1.8%). Most of the patients had one lesion (67.3%) and the size of many ulcers was 5-15 mm and most of them were in 4-6 cm of the anal margin. There were one ulcer in 13 cm and the other one in 20 cm of the anal margin. In the study by Madigan *et al*<sup>[7]</sup>, most of the patients had one ulcer (70%). The size of most of the ulcers was about 2 cm

and their distance from the anus was between 3 and 15 cm, but most of them were in 7-10 cm of the anal margin. It is stated in this study that there is one stage of disease in which there is no ulcer and it can be seen at the local inflammation of the rectum<sup>[7]</sup>. It seems that there were some patients in this stage that did not show any specific findings in colonoscopy or only showed redness and inflammation. In the study by Dehghani *et al.*<sup>[10]</sup> 11 out of 12 patients had between 1 to 4 superficial ulcers which was in 7 cm of rectosigmoid area and only 1 patient had polypoid lesion. In the study by Perito *et al.*<sup>[9]</sup> it is mentioned that the lesions of patients are mostly at the end part of rectum and in 10 cm of the anal margin. From 10 registered colonoscopy report, there were redness and inflammation in 8 patients and there was polypoid lesions in 4 of them.

According to the studies conducted so far, it seems that polypoid lesions are rare in the pediatric age group and most of the lesions are ulcers, erythema and inflammation; this has been reported in previous studies<sup>[14]</sup>.

All of the patients evaluated in this study, were initially treated with conservative treatments, recommendation to avoid straining during defecation and also dietary changes. Only the patients who did not respond to this method were treated with Sucralfate enema, Asacol suppository and also methylprednisolone injection around the lesion and finally 5 patients who did not also respond to these treatments, were treated with laparoscopic ventral rectopexy. During the follow-up, the symptoms of 69.8% of patients were recovered. Only 2 of these non-responsive patients were treated with rectopexy and the others with conservative treatments and use of Sucralfate enema and Asacol suppository.

In the study by Dehghani *et al.*<sup>[10]</sup> conservative treatments, and behavioral and dietary changes were recommended as the preliminary treatment. In that study, 58.3% of the patients (7 out of 12 patients) had the complete recovery of symptoms after treatment with Sucralfate enema and concluded that this is a suitable treatment for children. One of their patients responded to Salicylate enema, 1 to corticosteroid enema, 2 to corticosteroid injection and 1 of the patients were finally treated with rectopexy<sup>[10]</sup>.

In the study by Martín de Carpi *et al.*<sup>[15]</sup> which was conducted on 3 affected patients, 2 patients were treated with budesonide enema and 1 patient with only a dietary change. The symptom of all the 3 patients was recovered. In the study by Blackburn *et al.*<sup>[3]</sup> changing the behavior and encouraging children not to strain on defecation were recommended and the stool softeners were only used for the patients who had rigid stool; improvement was seen in all patients except for a patient who was not able to cooperate due to autism. This study indicated that most of the patients responded to behavioral change methods like biofeedback therapy<sup>[3]</sup>. In another study by Urgancı *et al.*<sup>[8]</sup> the patient's treatment began with Mesalazine enema, Sucralfate and

steroid enema.

These and other similar studies reveal that a comprehensive study has not been conducted so far to determine the best therapeutic procedures, so determining suitable treatment requires more and more complete examinations. But it seems that the treatment method without complications like trying to change diets and bowel habits is the best treatment in children; also, it is better to consider medical treatment and use of laxatives and enema with different substances as the second line of treatment<sup>[2,9]</sup>.

There are also different studies about the medical management in the children and most of the surgical cases have been conducted in patients with polypoid lesions or rectal prolapse and patients who were still symptomatic after trying several medical treatment<sup>[14,16,17]</sup>.

Bonnard *et al.*<sup>[17]</sup> have published the first successful rectopexy with laparoscopic method in a 12-year-old child who became asymptomatic in the next follow up and his colonoscopy became normal. In Godbole *et al.*<sup>[16]</sup> study, polypoid lesions and hidden rectal prolapse were diagnosed in the examinations for 1 of the 2 studied patients. Polypectomy through the anus and then ablation of the remained granulation were performed for this patient. The second patient had a large prolapse and was treated with rectopexy and the symptoms of both patients were recovered in the next follow-ups.

According to this and other similar studies, it seems that this disorder is not so rare in children in spite of what had been said about its rarity before and the reason of its low reporting is low familiarity of physicians with this disorder and its similarity with other common diseases of the anal canal and rectosigmoid. Therefore, the physicians should be aware of this disorder and thus prevent the late diagnosis of the disease and prevent its long term complications.

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## COMMENTS

### Background

Solitary rectal ulcer is defined as an infrequent but benign disorder of rectal and sigmoid colon which is diagnosed on the basis of clinical symptoms and histologic findings. The clinical symptoms in children are similar to adults, but since this disorder has not been well known in children, their symptoms can be confused with more common diseases of the rectum and sigmoid. It is very important to differentiate solitary rectal ulcer from other disorders of the rectum and sigmoid before starting treatment. Follow-up of these patients is necessary

in order to improve and sustain treatment method to prevent morbidity and recurrences which lead to the disease progresses to adulthood.

### Research frontiers

Solitary rectal ulcer is diagnosed as a cause of rectal bleeding in children in Pediatric Gastroenterology Section of Nemazee Hospital affiliated to Shiraz University of Medical Sciences and its frequency become increasing during the last years. However, there are very few English language literatures sources from Iran and other countries concerning the diagnosis and treatments of solitary rectal ulcer in children. The research hotspot is to introduce these real things happening to this population and to help other peers understand these backgrounds and trends in Iran.

### Innovations and breakthroughs

In recent years, the number of children with rectal bleeding who diagnosed as of solitary rectal ulcer has been increasing in Shiraz, Iran. The present study represents the largest pediatric series of solitary rectal ulcer in the world. On the other hand, the current data also suggested that this disorder is not so rare in children in spite of what had been said about its rarity before and the reason of its low reporting is low familiarity of physicians with this disorder and its similarity with other common diseases of the rectosigmoid. Therefore, the physicians should be aware of this disorder and thus prevent the late diagnosis of the disease and prevent its long term complications.

### Applications

The data in this study suggested that solitary rectal ulcer is not so rare in children and conservative and medical management for solitary rectal ulcer could yield relatively favorable outcomes. Furthermore, this study also provided readers with important information regarding the clinical and colonoscopic findings in these patients.

### Terminology

Solitary rectal ulcer is a benign and chronic disorder well known in young adults and less in children. It is often related to prolonged excessive straining or abnormal defecation and clinically presents as rectal bleeding, copious mucus discharge, feeling of incomplete defecation, and rarely rectal prolapse. Solitary rectal ulcer is diagnosed based on clinical symptoms and endoscopic and histological findings. The current treatments are suboptimal.

### Peer-review

Available papers concerning pediatric solitary rectal ulcer are scarce. The authors in this study analyzed the characteristics and outcomes of children with solitary rectal ulcer based on a large single-center series. This study showed that solitary rectal ulcer is not so rare in children. The results were interesting and provided important information concerning the background and trends of various treatments for solitary rectal ulcer in children.

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