

Massive lower gastrointestinal bleeding associated with solitary rectal ulcer in a patient with Behçet's disease

C. Bes¹, Ü. Dağlı², F. Yılmaz³, M. Soy⁴

¹Department of Rheumatology, Bakırköy Dr. Sadi Konuk Training and Research Hospital, Turkey;

²Department of Gastroenterology, Başkent University, Turkey; ³Department of Pathology, Abant İzzet Baysal University, Turkey; ⁴Department of Rheumatology, Hisar Intercontinental Hospital, Turkey

SUMMARY

Solitary rectal ulcer syndrome is a rare benign disorder that has a wide range of clinical presentations and variable endoscopic findings which makes it difficult to diagnose and treat. The clinical and endoscopic picture in this condition can also mimic malign ulceration, malignancy or Crohn's disease. Behçet's disease can affect the gastrointestinal tract. However to the best of our knowledge, no case with solitary rectal ulceration has been reported so far in literature. We herein present a patient diagnosed with Behçet's disease admitted to our clinic with rectal bleeding due to solitary rectal ulceration.

Key words: Solitary rectal ulcer; gastrointestinal bleeding; Behçet's disease.

Reumatismo, 2015; 67 (2): 82-84

■ INTRODUCTION

Solitary rectal ulcer syndrome (SRUS) is an uncommon clinical condition which presents a symptom complex of rectal bleeding, painful and mucous defecation, tenesmus, constipation and rectal prolapse. Although it was first identified in 1829 by Cruveilheir (1), it gained importance after its histopathological and clinical determination in 1969 by Madigan and Morson (2). Its etiology is still unclear, but it seems to be associated with trauma and local ischemia secondary to constipation, forced defecation and digital stimulation. Behçet's disease (BD) is a multisystem disorder associated with a triad of symptoms: recurrent oral aphtha, genital ulcer and uveitis. It can affect any part of the gastrointestinal tract, but it most often involves terminal ileum and caecum (3, 4). Unlike in Far Eastern countries, gastrointestinal involvement is not common in our country (5). To the best of our knowledge, the co-existence of BD and SRUS has never been

reported in literature. Herein we describe a case of BD, presenting with massive lower gastrointestinal bleeding associated with SRUS.

■ CASE REPORT

A 39 year-old man with Behçet's disease, followed up for seven years, was admitted to our outpatient clinic with rectal bleeding. He had ongoing rectal bleeding requiring a transfusion of 7 units. He had a history of recurrent aphthous stomatitis, scrotal ulcers, uveitis, thrombophlebitis, pulmonary embolism and neurological involvement. He was administered 100 mg/day azathioprine, 5 mg/day warfarin and 4 mg/day prednisolone. He was suffering from constipation and had a history of forced defecation (digital stimulation). An irregular broad hard lesion was palpated in the rectum about 5-6 cm from the anal verge. Colonoscopy revealed an abnormal map shaped ulceration extending 7-8 cm into the rectum from the beginning of anal

Corresponding author:

Cemal Bes

Department of Rheumatology, Bakırköy
Dr. Sadi Konuk Training
and Research Hospital, Turkey
E-mail: cemalbes@hotmail.com

canal (Figure 1). The histopathological analysis of biopsy specimens taken during colonoscopy showed an infiltration of mixed inflammatory cells with increased dysplastic tissue, cumulative gland units with an increased chromatin and mitotic activity (Figure 2). These findings were compatible with a solitary rectal ulcer. He was then diagnosed with SRUS and enema, and prednisolone was added to his treatment schedule. After this treatment rectal bleeding stopped and symptoms improved.

DISCUSSION

The solitary rectal ulcer syndrome is a benign rectal lesion most frequently seen in adults between 30-50 years of age. The most frequent symptom is rectal bleeding with a 90% incidence. Other symptoms are tenesmus, constipation, mucous defecation and lower quadrant stomach pain. Almost 25% of these subjects complain no symptoms.

On examination the rectum is perceived as thick and rigid or a polypoid mass may be detected (6). A typical colonoscopy finding of SRUS is an ulceration often located at 5-10 cm from the anal canal in the anterior rectal wall, surrounded with hyperemic, edematous mucosa with regular edges (7). SRUS can mimic inflammatory bowel diseases, ischemic colitis, cancer and trauma associated intussusception or rectal prolapse, therefore it can be potentially misdiagnosed. Due to its complex clinical and endoscopic findings, a biopsy and an histopathological examination are required to confirm the diagnosis. The main pathological findings are fibromuscular dysplasia, collagenous and muscle cell infiltration in the lamina propria, a thickening of muscularis mucosa and deformations in crypt units (8).

Although there are various treatment options that range from conservative therapy to surgery, so far no therapy has proved to be more effective than the others. Surgery, medical therapy or biofeedback can be applied when conservative therapy is not helpful. The medical therapy consists of the following options: 5-aminosalicylic

acid enema, sucralfate enema and steroid enema applications (9, 10). The selection and success of therapy varies from patient to patient. Recently, the argon plasma coagulation technique has been reported to be effective in SRUS therapy (11). Patients

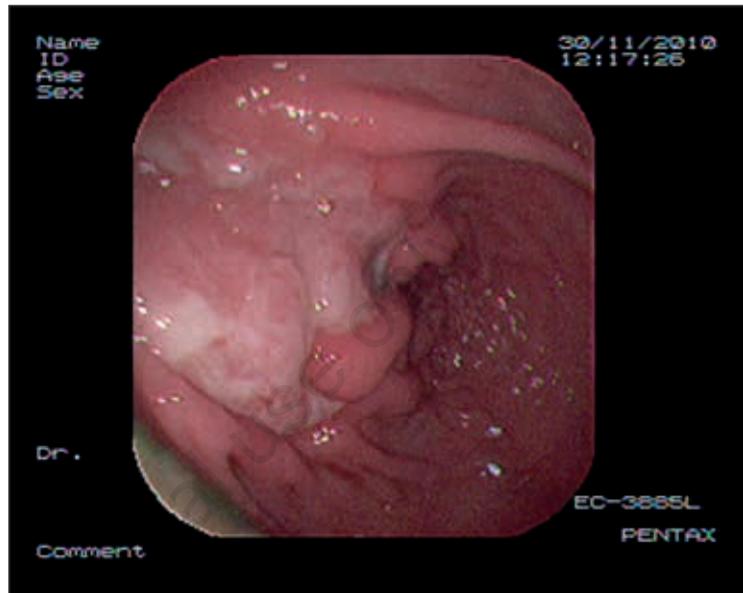


Figure 1 - Colonoscopy: shaped ulceration and surrounding mucosal erythema.

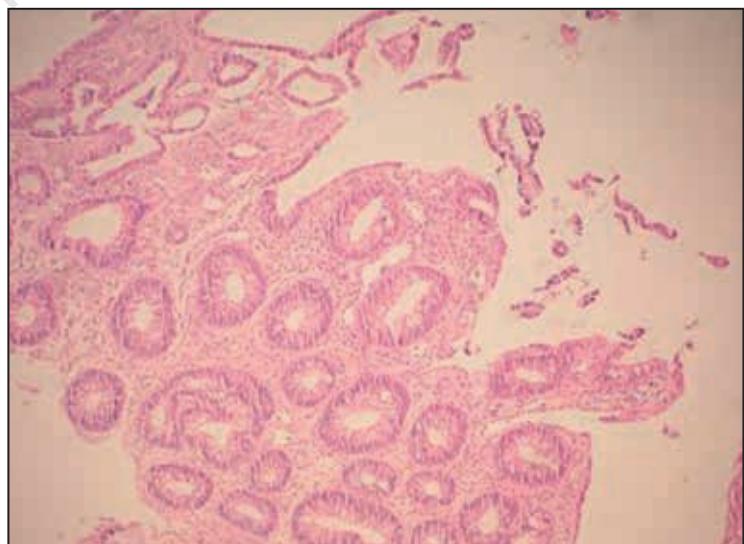


Figure 2 - Histopathological examination: the rectal mucosa shows an infiltration of mixed inflammatory cells with increased dysplastic tissue, cumulative gland units with increased chromatin and mitotic activity (HE stain $\times 40$).

should also be discouraged from using the digital stimulation technique to ease defecation. Gastrointestinal involvement is detected in more than 40% of patients with Behçet's disease (12). Gastrointestinal symptoms are stomachache, diarrhea, nausea and abdominal distention. Although gastrointestinal symptoms are common in BD, gastrointestinal ulcer (intestinal BD) has a 1-2% incidence (13).

The most common sites of ulceration are the terminal ileum and the caecum, yet any area of gastrointestinal system can be involved. Only 15% of cases present a diffuse colon involvement in which rectum is beware. Behçet's colitis doesn't affect the rectum like Crohn's disease. Ulcerations are generally large bottomed and separated from each other and might be located in normal or inflamed mucosa (14). The histopathological examination of the lesion may reveal a vasculitic innovation in small and middle sized vessels. A dense infiltration of lymphocytes can be determined in the perivascular area (15). Rectal involvement is not common in BD. SRUS must be suspected in patients diagnosed with Behçet's disease affected by massive rectal bleeding. In conclusion, this is the first study reporting the coexistence of Behçet's disease and SRUS in literature. Symptoms including rectal bleeding, tenesmus and mucous defecation SRUS must be taken into consideration for the purpose of differential diagnosis.

■ REFERENCES

1. Cruveilhier J. Ulcere chronique du rectum. Anatomie pathologique du corps human. Paris: JB Billiere; 1829.
2. Madigan MR, Morson BC. Solitary ulcer of the rectum. Gut. 1969; 10: 871-81.
3. da Fonseca LM, Souza Fde L, Arantes V, da Silva RG. Giant refractory solitary rectal ulcer syndrome treated with topical sucralfate. Int J Colorectal Dis. 2010; 25: 1025-6.
4. Kasahara Y, Tanaka S, Nishino M, Umemura H, Shiraha S, Kuyama T. Intestinal involvement in Behçet's disease: review of 136 surgical cases in the Japanese literature. Dis Colon Rectum. 1981; 24: 103-6.
5. Yurdakul S, Tüzüner N, Yurdakul I, Hamuryudan V, Yazici H. Gastrointestinal involvement in Behçet's syndrome: a controlled study. Ann Rheum Dis. 1996; 55: 208-10.
6. Saadah OI, Al-Hubayshi MS, Ghanem AT. Solitary rectal ulcer syndrome presenting as polypoid mass lesions in a young girl. World J Gastrointest Oncol. 2010; 2: 332-4.
7. Shara Al, Azar C, Amr SS, Haddad M, Eloubeidi MA. Solitary rectal ulcer syndrome: endoscopic spectrum and review of the literature. Gastrointest Endosc. 2005; 62: 755-62.
8. Remes-Troche JM, Rao S SC. Anorectal motor disorders. Best Pract Res Clin Gastroenterol. 2007; 21: 733-48.
9. Bishop PR, Nowicki MJ. Nonsurgical therapy for solitary rectal ulcer syndrome. Curr Treat Options Gastroenterol. 2002; 5: 212-23.
10. Zargar SA, Khuroo MS, Mahajan R. Sucralfate retention enemas in solitary rectal ulcer syndrome. Dis Colon Rectum. 1991; 34: 455-7.
11. Somani SK, Ghosh A, Avasthi G, Goyal R, Gupta P. Healing of solitary rectal ulcers with multiple sessions of argon plasma coagulation. Dig Endosc. 2010; 22: 107-11.
12. Oshima Y, Shimizu T, Yokohari R, Matsumoto T, Kano K, Kagmani T, et al. Clinical studies on Behçet's syndrome. Ann Rheum Dis. 1963; 22: 36-45.
13. Masugi J, Matsui T, Fujimori T, Maeda S. A case of Behçet's disease with multiple longitudinal ulcers all over the colon. Am J Gastroenterol. 1994; 89: 778-80.
14. Smith JA, Siddiqui D. Intestinal Behçet's disease presenting as a massive acute lower gastrointestinal bleeding. Dig Dis Sci. 2002; 47: 517-21.
15. Lee RG. The colitis of Behçet's syndrome. Am J Surg Pathol. 1986; 10: 888-93.