Solitary Rectal Ulcer Mimicking a Malignant Stricture.  
A Case Report

Evangelos Perrakis¹, Antonios Vezakis¹, Georgios Velimezis¹, Dimitrios Filippou²

1) Department of Surgery, Western Attica General Hospital, Athens. 2) 1st Department of General Surgery, Pireaus GP Hospital “Tzaneio”, Greece

Abstract
Solitary rectal ulcer syndrome is a rare disorder. The macroscopic appearance varies from single to multiple ulcers or even circumferential lesions. A patient is presented with solitary rectal ulcer syndrome causing circumferential stenosis of the rectum. Solitary rectal ulcer syndrome should be considered in all patients with malignant looking rectal tumours. Surgical resection is usually required in patients with rectal stricture. This can be performed either through a laparotomy or a parasacral transsphincteric approach.

Key words
Solitary rectal ulcer - surgical resection - parasacral approach

Rezumat
Ulcerul rectal solitar este o afecțiune rară. Aspectul macroscopic este variabil de la ulcere unice sau multiple, până la leziuni circumferențiale. Este prezentat un pacient cu ulcer rectal solitar, care a produs o stenoză circumferențială a rectului. Ulcerul rectal solitar trebuie luat în considerare la toți pacienții cu tumori rectale aparent maligne. Rezecția chirurgicală este necesară în prezența stenozei rectale. Aceasta poate fi efectuată fie prin laparotomie fie prin abord transsfincterian parasacral.

Rezumat
Solitary rectal ulcer syndrome is a rare disorder. The macroscopic appearance varies from single to multiple ulcers or even circumferential lesions. A patient is presented with solitary rectal ulcer syndrome causing circumferential stenosis of the rectum. Solitary rectal ulcer syndrome should be considered in all patients with malignant looking rectal tumours. Surgical resection is usually required in patients with rectal stricture. This can be performed either through a laparotomy or a parasacral transsphincteric approach.

Case report
A 52 year old lady presented with altered bowel habits (constipation) and a feeling of inadequate defecation. She had no history of constipation in the past or excessive straining at stools. She had been a regular user of analgesic suppositories (paracetamol 400 mg plus codeine 20 mg plus caffeine 50mg) for the relief of migraine during the last year.

Rectal examination revealed a hard ulcerated lesion at 3 cm on the posterior rectal wall occupying nearly half the circumference of the rectum. The lesion was thought to be malignant, biopsies were taken and the patient was placed on the list for an abdominoperineal resection. There was no evidence of visible rectal prolapse, while straining in the squatting position. Surprisingly, the biopsies showed chronic inflammation with fibrous obliteration of the lamina propria without any evidence of malignancy. The patient had an MRI scan, which showed a thickened and oedematous rectal wall with no evidence of extramural spread, and she was discharged.

She was re-admitted a month later for examination of the rectum under anaesthesia. This revealed a hard ulcerated lesion occupying the whole circumference of the rectal wall and causing a stricture (Fig.1), which allowed only a black Nelaton catheter CH 10 (Maersk Medical A/S) to pass through. Biopsies were taken, and the next day the patient had a laparoscopy and a right transverse loop colostomy.

In conclusion, surgical resection was performed through a transsphincteric parasacral approach.
with an end to end anastomosis. Recovery was uneventful. The colostomy was closed two months later. Two years after her second operation the patient remains asymptomatic, with no ulcer seen at endoscopy (Fig.2) and with normal defecation. The histology of the specimen showed chronic inflammation with replacement of the lamina propria with fibroblasts and smooth muscle cells.

Discussion

Solitary rectal ulcer syndrome is a chronic disorder. The mean age at presentation is the third and fourth decade with a range from 10 to more than 80 years. There is a slight female predominance (4). The usual presenting symptoms are the passage of mucus and blood, constipation, tenesmus, anorectal pain, a feeling of incomplete evacuation and straining at defecation. In one series (4) rectal bleeding was documented in 56%, straining in 28%, passage of mucus in 18%, constipation or diarrhoea in 55%, and 26% of patients were asymptomatic. Our patient presented with altered bowel habits (constipation) and a feeling of incomplete evacuation.

Solitary rectal ulcer syndrome is a rare disease, the symptoms are not characteristic and it is usually misdiagnosed as inflammatory bowel disease or malignancy. Our first clinical diagnosis was rectal cancer. Biopsies are essential in excluding malignancy.

The histologic features of solitary rectal ulcer syndrome are well documented. The lamina propria is replaced by fibroblasts, smooth muscle and collagen, with associated hyper trophy and disorganisation of the muscularis mucosa (2). The lesion may appear macroscopically as erythema, single ulcer, multiple ulcers or polypoid lesion. In one series the reported prevalence of ulceration was 57%, of polypoid lesions 25% and of patches of hyperaemic mucosa in 18% of patients (3). Lesions were multiple in 30% of cases in another series (2). Usually, they appear on the anterior rectal wall (68 - 95% of cases) approximately 6 to 10 cm from the anal verge (4-6). In our case the ulceration was very low at 3 cm, starting from the posterior rectal wall, where it soon became circumferential.

The pathogenesis of solitary rectal ulcer syndrome remains obscure. Self digitation has been suggested as a cause (7), but the lesions are often located beyond the reach of a finger. Prolapse induced by rectal mucosal trauma or ischaemia has been strongly implicated as a possible cause. The incidence of rectal prolapse (either mucosal or full rectal) in patients with solitary rectal ulcer syndrome varies from 13 to 94% (4). A combination of rectal prolapse and paradoxical contraction of the pelvic floor have been suggested to cause local ischaemic injury and trauma to the rectal mucosa, resulting in ulceration. The anterior wall mucosa 7 to 10 cm proximal to the anal verge is often the first and largest part of the intussusceptum, as it descends downwards into the anal canal. This corresponds to the usual location of ulcerations seen clinically. However, electromyography has failed to find evidence of a hyperactive rectal sphincter in the majority of patients (4). The possible role of ischaemia in the pathogenesis of solitary rectal ulcer syndrome is further supported by the association between suppositories of ergotamine (an α-adrenoreceptor agonist with potent vasoconstrictor action) and development of the syndrome (9). Psychological problems expressed as a disturbance in toileting behaviour have been suggested as an important pathogenetic factor in some patients. The improvement of symptoms with behavioural treatment (biofeedback) supports this as an important factor (10,11).

Our patient had no clinical evidence of rectal prolapse. Trauma to the rectal mucosa by the suppositories could have been the cause. This is supported by the location of the ulcer to the posterior rectal wall and the low position at 3 cm, where the injury caused by suppositories is likely to occur. Surprisingly, the ulcer had extended very quickly to cause a circumferential stricture of the rectum. Steroid enemas were not effective, as has been proposed by others (3).

Surgery is advised for symptomatic solitary rectal ulcer syndrome patients with rectal prolapse because of the poor
response to medical therapy. The procedure of choice in patients with rectal prolapse is abdominal rectopexy (12). In non prolapsing patients with intractable symptoms, local excision, colonic resection or diverting colostomy are required. In one study, resection and coloanal anastomosis for benign rectal lesions in two patients with solitary rectal ulcer syndrome had poor results, both requiring a colostomy (13). Resection of the involved part of the rectum via a parasacral transsphincteric approach is an alternative to low anterior resection especially for very low rectal lesions. It permits good access to the lower rectum and laparotomy is avoided. In our case it was preceded by a laparoscopic right transverse loop colostomy to avoid complete bowel obstruction.

Conclusions

In conclusion, symptomatic solitary rectal ulcer syndrome should always be considered in patients with malignant looking rectal tumours. In cases of circumferential lesions causing stenosis of the rectum, surgical resection is required. The parasacral approach remains an alternative to low anterior resection for very low situated lesions.

References