

Case report

Aphthous colitis in a woman with diverticulitis

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SUMMARY

Diverticulitis is a well-established complication of diverticulosis. Data regarding the endoscopic appearance of diverticulitis are limited, as colonoscopy is generally contraindicated during the acute attack, due to the increased risk of complications, especially bowel perforation. Endoscopic signs of inflammation in patients with diverticulosis are limited to the segments having the diverticulae, with rectal sparing, and appear in the literature as segmental colitis.

We report a case of a middle-aged woman with diverticulitis who developed aphthous lesions in the rectum and sigmoid a few days after the initiation of symptoms. Other causes of aphthous lesions of the colonic mucosa (ie. Behcet's disease, Crohn, ischemia, tuberculosis etc.) were excluded. We suggest that the presence of aphthous lesions should not be included in the setting of segmental colitis, but should be considered as endoscopic signs of diverticulitis, possibly caused by inflammation and ischemia.

Key words: Aphthae, aphthous colitis, diverticulitis, segmental colitis associated with diverticulosis, Crohn's disease.

INTRODUCTION

Diverticulitis is a well-established complication of diverticulosis. Most patients are over 50 years of age and present with abdominal pain, fever and diarrhea or constipation. On clinical examination there is tenderness, usually on the left lower abdominal quadrant, with or without rebound tenderness. An episode of diverticulitis is possibly the end point of macro- or microperforation

of a diverticulum. Acute perforation follows an abrupt increase in the endoluminal pressure, while microperforation could be related to the formation of a fecalith (a firm mass of undigested fiber and bacteria) in the diverticulum, which presses the sac and causes inflammation, erosion, and microperforation.

Data on the endoscopic appearance of acute diverticulitis are very limited. The reason is that endoscopy is generally avoided, due to the increased risk of perforation. Segmental colitis in patients with diverticulosis (SCAD) has been well known for more than two decades.^{1,2} Two recent studies deal with the incidence, clinical picture and outcome of patients with SCAD^{3,4}. The condition resembles inflammatory bowel disease, mainly Crohn's, with patchy distribution and rectal sparing.

Aphthous colitis has been attributed to a variety of clinical entities. Aphthae and focal inflammation with normal surrounding mucosa can be found in an early stage of Crohn's disease.^{5,6} It may also be noticed in Behcet's disease in conjunction with oral genital ulcers and uveitis. Aphthous lesions have also been reported in bowel ischemia and in the early phase of intestinal tuberculosis.⁷

Aphthous ulcers in conjunction with diverticulitis have been described recently in a case report by Jung et al.⁸ In the past, this association was reported only in studies of surgical specimens.^{9,10,11} We report a case of a woman with diverticulitis who presented with aphthous colonic lesions.

CASE REPORT

A 51-year-old female nurse was admitted to the Department of Surgery with acute left abdominal pain, fever and constipation. She had no significant past medical history and had not taken any drugs. On clinical examination there was diffuse abdominal tenderness, especially in the left lower quadrant and mild rebound tenderness.

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The white cell blood count was elevated at $12200/\text{mm}^3$ ($5.000\text{-}10000/\text{mm}^3$), ESR:50/1h, CRP: 90 mg/l (< 5 mg/dl). Serum electrolytes, renal and liver function tests were normal and stool culture and parasitology were also negative. She was treated with antibiotics (ciprofloxacin 400 mgx2 and metronidazole 500 mgx3 IV for 8 days). The patient became afebrile two days after the initiation of treatment.

We were asked to perform a colonoscopy, after preparation with enemas, three days after admission. It was a total colonoscopy and revealed multiple diverticulae with some macroscopic signs of mucosal inflammation (especially hyperemia) in the sigmoid colon (Figure 1) and a protruding mass with central umbilication and ulceration at 28 cm from the anus (Figure 2). One week later, we performed a sigmoidoscopy to further define

the exact nature of the mass. After taking the first biopsy specimen, an amount of pus appeared in the lumen (Figure 3). Microbiologists cultured *E. Coli* and *Proteus*. We also noticed, for the first time, the presence of multiple aphthous ulcers throughout the examined segments (rectum and sigmoid) (Figure 4). There were no yellow membranes suggesting pseudomembranous colitis, exudates or any specific macroscopic evidence of inflammatory bowel disease. Histologic examination showed moderate to severe inflammatory infiltrate with small necrotic areas and microabscesses. There was also slight distortion of the glandular architecture and focal apoptosis of the surface epithelium. These features were considered compatible with infectious colitis, although inflammatory bowel disease could not be totally excluded. On day 17, the patient was discharged in good health.

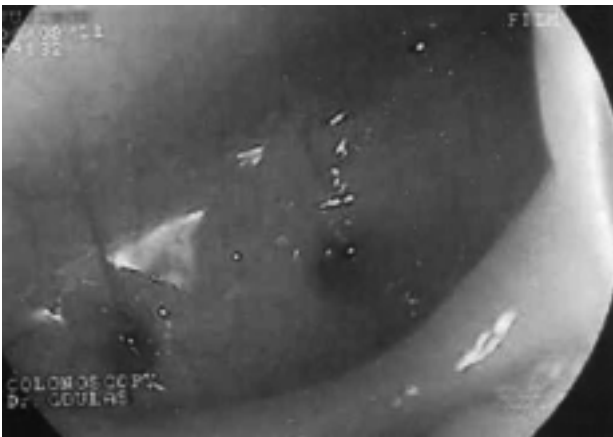


Fig. 1 Multiple diverticulae of the sigmoid colon.

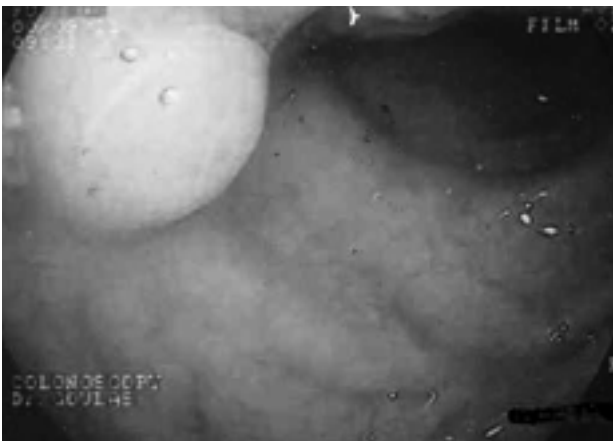


Fig. 2 The protruding mass with superficial ulceration.

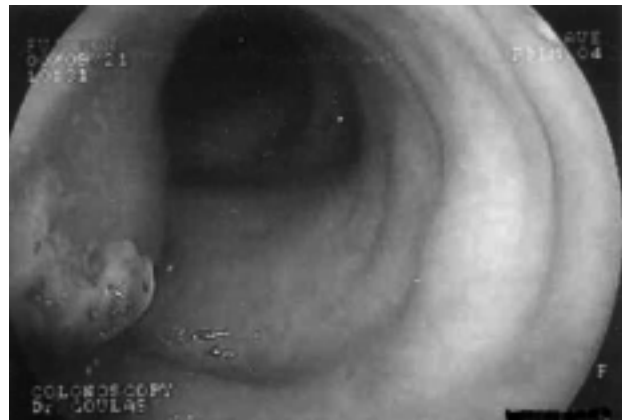


Fig. 3 Discharge of pus from the mass after taking the first biopsy (abscess).

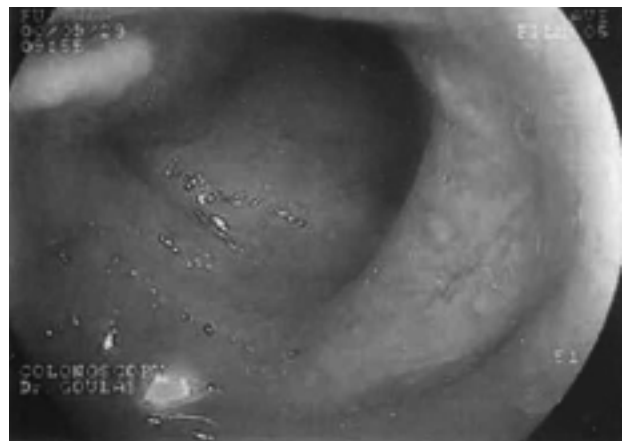


Fig. 4 Aphthous ulcers surrounded by red halo in the sigmoid colon.

Fifty days later, on follow-up endoscopy, multiple diverticulae and significant regression of the mass were noted. No aphthous lesions were present. At that time, the patient was asymptomatic. Eighteen months later, after a further attack of diverticulitis, that ceased conservatively, she had a left hemicolectomy. The surgical specimen macroscopically included multiple diverticulae and a benign stricture in the sigmoid but no microscopic evidence of Crohn's disease. At present, 4 years after the initial episode, the patient is doing well with no clinical recurrence and no evidence of IBD.

DISCUSSION

Segmental colitis in patients with diverticulosis (SCAD) has been described in the past.^{1,2,3,4} In a large recent prospective Italian study the criteria for diagnosing SCAD were edema, focal erythema, friability submucosal ecchymosis, erosions and ulcers in the diverticular tract of the colon and 14/5457 (0,25%) patients met these criteria.³ Among those patients, 13/14 presented with hematochezia, 7 with diarrhea and 5 with pain. None had fever. Stool examinations were negative while white blood count and CRP were normal. The histology revealed non-specific inflammation. According to the authors their clinical history and laboratory results did not fit the diagnosis of diverticulitis. Patients were treated with 5-ASA and were in remission after 6 weeks and one year suggesting a self limiting illness rather than IBD.

Early this year Koutroubakis et al, in a large retrospective study, identified 23/7391 (0,31%) cases of SCAD⁴. These represent a figure of 3,8% among patients with diverticulosis. Presenting symptoms were hematochezia in 18 cases, abdominal pain in 14 and diarrhea in 9. No patients had fever and blood tests were normal. In all patients lesions were found in the left colon, with rectal sparing while right colon involvement was noted in 2. Histology was characteristic of ulcerative colitis in 4 cases, ischemic in 1, SCAD in 6 while the remaining 12 had minimal lesions. On follow up, 4 patients were refractory to medical treatment (antibiotics +/- 5-ASA) and had a sigmoid colectomy with no recurrence. Of the remaining 19, 5 had episodes of relapse, which responded to medical treatment. The authors conclude that SCAD is a clinical entity distinct from IBD, with good response to medical treatment and no evolution to IBD in the long term.

In our case, the clinical setting was clearly that of acute diverticulitis and endoscopy revealed aphthous

colitis. The basic question is whether aphthous colitis is an endoscopic feature in the setting of diverticulitis or whether it results from another cause (IBD, ischemia etc.) In the past, 3 studies have failed to show coexistence of Crohn's disease and diverticulitis. In the first study none out of 8 patients with granulomatous colitis and diverticulitis developed Crohn's disease.⁹ In a second study no patients were diagnosed with Crohn's disease, out of 25 patients with diverticulitis and endoscopic lesions suggesting Crohn's colitis.¹⁰ In the third study, the authors studied resection specimens with macroscopic features of both diverticulitis and Crohn's and found that in the 9/11 cases, changes were confined to the segment having the diverticulae.¹¹ It must be emphasized that all 3 studies related to surgical specimens.

Recently, a case of aphthous colitis in a young man with diverticulitis was reported by Jung et al.⁸ Aphthous lesions were extended in the proximal large bowel, beyond a stricture and far away from the diverticular segment. They were most prominent in the cecum. The authors suggest that aphthous lesions in the colon may be part of the spectrum of mucosal changes of diverticulitis, secondary to inflammation, ischemia, bowel distention and infection.

Our case comes to reinforce this suggestion. The patient had an attack of acute diverticulitis complicated by an abscess formation and one week after the initiation of symptoms, multiple aphthous ulcers were detected in the rectum and the sigmoid colon. She was treated with antibiotics, recovered and the aphthous colitis disappeared. Crohn's disease, Behcet's disease, ischemia, infectious as well as other forms of colitis had been excluded. Finally, after a further episode of diverticulitis, she had a left colectomy and the surgical specimen revealed diverticulosis and a benign stricture but no evidence of Crohn's or any other kind of bowel disease. The extended follow up (4 years) free of recurrence established the exclusion of IBD.

The lack of other similar reports linking aphthous lesions to diverticulitis, except that of Jung et al,⁸ is obviously due to the fact that, in patients with diverticulitis, endoscopy is avoided due to the increased risk of complications. In both cases, the presence of aphthous lesions outside the diverticular segment may be a sequelae of diverticulitis. In our case, a complete recovery was achieved with antibiotics.

We conclude that aphthous lesions in the colon may be considered as an endoscopic sign of diverticulitis, probably caused by inflammation, local ischemia and infection.

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