

## A Case Report of Ulcerative Colitis Presenting with Perianal Disease in 78-Years-Old Female Patient

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### ABSTRACT

**Introduction:** IBD usually presents with different phenotypes, which may include the presence of perianal disease. Although perianal disease is typical for patients with Crohn's disease, we should always pay the attention to the possibility of perianal disease presence (hemorrhoids, anal fissure and/or fistula, perianal abscess) in patients with ulcerative colitis. In a certain number of patients with Crohn's disease, the perianal fistulas develop before the intestinal disease and the occurrence of perianal disease prior to development of ulcerative colitis is very rare.

**Case presentation:** We report a case of a 78-year-old female patient who was admitted to our hospital with the complaints of perianal pain, fever, rectal bleeding, abdominal cramps and progressively worsening bloody diarrhea. The results of diagnostic work-up was consistent with the diagnosis of ulcerative colitis and the presence of perianal disease in a form of anal fissure and rectocutaneous fistula. The treatment included the combination therapy of antibiotics, corticosteroids, 5-ASA compounds, and parenteral supplementation of packed erythrocytes, albumins and iron.

**Conclusion:** In presented older female patient with ulcerative colitis, the perianal disease was healed with combination therapy of antibiotics, corticosteroids and 5-ASA compounds, reflecting the difference in pathogenesis, clinical course and therapeutic response, compared to the nature of perianal disease in patients with Crohn's disease. The alternation of chronic constipation and diarrhea could be of pathogenetic influence in development of perianal disease, in this particular patient with ulcerative colitis.

### Keywords

Inflammatory bowel disease, Ulcerative colitis, Crohn's disease, Perianal disease, Anal fissure.

### Abbreviations

IBD: Inflammatory Bowel Disease; GI: Gastrointestinal; CD: Crohn disease; UC: Ulcerative Colitis; MRI: Magnetic Resonance Imaging; TPUS: Transperineal Ultrasonography, PAD: Perianal disease.

### Introduction

Inflammatory bowel diseases (IBDs) are chronic immune-mediated diseases of gastrointestinal (GI) tract with a periods of remission and relapse. A complex interaction between host immune system, environment and microbiota is crucial for development of IBD, in a genetic susceptible host. The intestinal dysbiosis may alter the mucosal immune response and the type of diet is considered as a risk factor for IBD development. Studies in humans have clearly shown the differences in composition and

abundance of gut microbiota in patients with IBD, compared to healthy controls [1,2].

IBD usually presents with different phenotypes, which may include the presence of perianal disease (PAD). Although PAD is typical for patients with Crohn's disease (CD), the attending gastroenterologist should always pay the attention to the possibility of PAD presence, such as hemorrhoids, anal fissure and/or fistula, perianal abscess in patients with ulcerative colitis (UC) [3]. The clinical course of CD with PAD, especially with perianal fistulas and abscesses is often characterized with more rapid progression to stricturing and/or penetrating complications. On the other hand, in patients with UC the intestinal inflammation is limited to the mucosa which raises the question of pathogenetic link of PAD with etiology of UC. Nevertheless, the presence of PAD in patients with UC is of clinical importance, causing anal pain and discomfort linked to defecation problems which significantly impairs the overall quality of life in these patients. Furthermore, the presence of PAD in UC is not such rare condition and it could represent a misleading problem in clinical practice, sometimes being confused with the relapse of UC or raising the question of misdiagnosed CD [4].

The authors present a case of 78-years old female patient with UC and PAD, discussing the diagnosis, epidemiology and clinical impact of PAD in patients with UC.

### Case Presentation

We report a case of a 78-year-old female patient who was admitted to our hospital with the complaints of perianal pain, fever, rectal bleeding, abdominal cramps and progressively worsening bloody diarrhea. The patient has been immobile due to longstanding musculoskeletal condition and has been diagnosed with ulcerative proctitis, two years before the actual admission. During that period of two years the patient experienced the alternating constipation and bloody diarrhea, occasionally. The previous proctologic examination, performed two years before actual admission showed the presence of anal pressure ulcer, anal fissure and erosions of the rectal mucosa on anorectoscopy.

On actual admission, the physical examination observed a linear to ovoid shaped ulcerating and bleeding anal fissure. (Figure 1) Upon admission, the diagnostic work-up included laboratory testing and stool studies, upper and lower GI endoscopy with histology, small bowel videoendocapsule and cross-sectional radiologic imaging.



**Figure 1:** Picture is presenting anal fissure, perianal inflammation and a moisture lesion on admission day.

The laboratory data showed microcytic hypochromic anemia, iron deficiency, normal total leukocyte and platelet count, elevated erythrocyte sedimentation rate (ESR) (45 mm/hr) and elevated CRP (165 mg/L). The stool studies, including *C difficile* toxin A/B assay were negative.

Esophagogastroduodenoscopy with biopsy showed chronic gastritis and testing for *H. pylori* infection was negative. Total colonoscopy with terminal ileoscopy showed diffusely hyperemic and edematous mucosa of the rectum, sigmoid and descending colon, with disappearance of the submucosal vascular pattern, granular appearance of the mucosa, and few scattered shallow ulcerations with fibrin and clots. Pathology revealed the inflammation limited to the mucosa, characterized with predominantly neutrophilic infiltrate and crypt abscesses.

Multislice computerized tomography (MSCT) showed significant wall thickening from left colic flexure in continuity to the rectum, and in right anal region anal fistula leading to the skin surface (Figure 2). Videoendocapsule investigation did not detect any inflammatory changes in the small bowel. The results of current endoscopy with histology, as well as the findings of cross-sectional radiology imaging were consistent with diagnostic criteria of UC associated with PAD, in a form of anal fissure and perianal fistula.



**Figure 2:** MSCT image is presenting rectal wall thickening, blurring of perirectal fat tissue due to inflammation, rectocutaneous fistula on the right side of the pelvis.

The treatment included a combination of antibiotics (ciprofloxacin and metronidazole), transfusion of packed erythrocytes, parenteral supplementation of albumins and iron. The therapeutic approach also included corticosteroids and 5-ASA compounds. After two weeks of treatment, the patient indicated the subjective improvement and the regression of anal fissure was also noticed. One month later, the diarrhea has ceased and there was no blood in the stool. The anal fissure was significantly smaller and the results of control laboratory investigation were within normal range. The patient continued the therapy with 5-ASA, orally and topically.



**Figure 3:** Picture is presenting closed anal fissure, perianal inflammation and a moisture lesion healed after treatment.

The repeated lower-GI endoscopy showed the improvement of inflammatory lesions in the left colon and the rectum. Two years after the discharge, the patient underwent a MSCT scan that revealed normal thickness of the rectal wall with no signs of perirectal fat tissue inflammation and no signs of rectocutaneous fistula.

### Discussion

This report revealed the case of UC associated with PAD in a form of anal fissure and perianal fistula, in an older female patient. The fact that PAD healed after the initial therapy with antibiotics and corticosteroids, in combination with 5-ASA as a form of long-term therapy speaks in favour of pathogenetic influence of alternation of chronic constipation and diarrhea in development of PAD, in this particular patient with UC. On the other hand, PAD in CD represents the phenotype of the disease, caused by the pathogenetic mechanisms that characterize the clinical course of CD.

The medical history and physical examination which includes anorectal examination represent the first step in diagnosis of PAD [4-7]. The results of laboratory testing, the findings of upper and lower GI endoscopy and cross-sectional imaging can differentiate between CD and UC in majority of cases. Magnetic resonance imaging (MRI) and transperineal ultrasonography (TPUS) are radiological techniques useful for evaluation of perianal disease in patients with IBD. A pelvic MRI is used as first diagnostic procedure, but TPUS is cheaper and it can be used to follow-up patients with perianal disease [8]. Endoscopic ultrasound (EUS) can be useful for showing the structure of the sphincter and pelvic MRI is gold standard for evaluation of perianal fistula, it is noninvasive and can also detect silent abscesses and inflammation [4]. However, in older and immobile patient, the use of multiple treatment modalities may be associated with difficulties in overall patient care and, using the single method of cross-sectional radiologic imaging, such as MSCT in combination with endoscopy and videocapsule we obtained sufficient information

for the diagnosis of UC, associated with non-complicated PAD in the presented patient. However, fistulizing perianal PAD is quite common in patients with CD and when fistulas occur in the setting of UC, the diagnosis is often questioned [9-11].

The reported incidence of perianal fistula in CD ranges from 17% up to 50% [11], and the incidence of anal fissures is up to 27.6% [12]. In approximately 10% of patients, perianal fistulization is the initial manifestation of Crohn's disease [3]. The data in the literature speak of PAD incidence in UC patients in up to 5%, being more frequent in men, in patients with distal disease and/or extensive disease [10,13,14]. It is important to emphasize that the change in diagnosis (UC to CD) occurs in a certain number of patients, mainly due to transmural involvement in colectomy specimen, small intestinal involvement in later course of disease and/or change in endoscopic appearance of intestinal mucosa on repeated endoscopies [14]. The change in diagnosis occurs in approximately one-third of patients with UC, suffering with complex PAD and it does not occur in any patient with simple PAD [15]. In addition, we did not find any data in published literature on PAD, occurring before the clinically overt picture of UC. In the presented patient, PAD was documented before the full clinical picture of UC was documented and the fistula was noted at the same time when diagnosis of UC was reached.

A recent retrospective study reviewed the data on 944 patients with initial diagnosis of UC during the follow-up period to 10 years [16]. In this study, the authors have reported the cumulative incidence of PAD (bleeding hemorrhoids, anal fissures, abscesses and fistulae) of 8.1% and 16.0% at 5 and 10 years respectively. The cumulative incidence of anal fissures was reported at 10 years in 5.3% of the patients included in the analysis. The authors of the mentioned study concluded that a sizeable proportion of UC patients actually suffer from anal problems and hypothesized that the higher rate of anal problems in UC patients compared to the general population could be due to chronic and recurrent diarrhea [16]. This is consistent with the influence of alternation of chronic constipation and diarrhea in development of PAD, in the case of presented patient with UC.

Another cross-sectional study, based on returned validated questionnaires that were sent to the members of Dutch National Crohn's and Colitis patient organisation reported the incidence of active PAD in 39% of patients with CD, 16% of UC patients (of which 84% anal fissures) and in 20% of unclassifiable IBD patients [17]. In the mentioned community-based IBD population, the study has shown that patients with PAD more frequently reported the symptoms of fecal incontinence, diminished quality of life and a higher rate of disability, as a result of their disease. This observation also implies that PAD may be the cause of significant burden for the healthcare resources [18].

### Conclusion

In clinical routine, the physician should always pay the attention to the possibility of PAD presence, such as hemorrhoids, anal fissure and/or fistula and perianal abscess in patients with UC. In

a certain number of patients with CD, PAD develops before the intestinal disease and the occurrence of perianal disease prior to development of ulcerative colitis is very rare. In UC patients, the occurrence of PAD (predominantly anal fissures) has a different pathogenetic background than PAD in CD patients, although the change in diagnosis (UC to CD) occurs in a certain number of patients with UC, mainly suffering with complex PAD. The presence of PAD in UC patients has a negative influence on quality of life and may be the cause of increased use of healthcare resources. In presented older female patient with UC, PAD was healed with combination therapy of antibiotics, corticosteroids and 5-ASA compounds, reflecting the difference in pathogenesis, clinical course and therapeutic response, compared to the nature of PAD in CD patients.

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### Ethical Statement

The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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