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ULCERATIVE COLITIS COMPLICATED BY COLOCUTANEUS FISTULA: A CASE STUDY

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Background. Ulcerative colitis (UC) is a multifactorial chronic inflammatory bowel disease characterized by such symptoms as abdominal pain, diarrhea, and rectal bleeding. One of the complications of UC is the formation of fistulas with the bladder, vagina, perineum, and other structures.

Objective. To present a clinical case of ulcerative colitis complicated by colocutaneous fistula.

Material and Methods. Clinical observation data of a 68-year-old man having been treated in the Purulent Surgery Department of the Grodno University Clinic.

Results. The patient, with a low body mass index (17.1 kg/m²), was admitted for leakage of intestinal contents from postoperative scar (with a history of a midline laparotomy for intestinal obstruction with creation of a loop ileostomy). Imaging confirmed a fistula to the sigmoid colon, and colonoscopy revealed erosive proctosigmoiditis. A left-sided hemicolectomy was performed. Histological findings were consistent with ulcerative colitis. Postoperatively, an abdominal abscess developed, which was successfully treated with drainage.

Conclusion. The case illustrates the complexities of UC management and its multifactorial nature.

Keywords: *ulcerative colitis, inflammatory bowel disease, colonocutaneous fistula.*

КЛИНИЧЕСКИЙ СЛУЧАЙ ЯЗВЕННОГО КОЛИТА, ОСЛОЖНЕННОГО КИШЕЧНО-КОЖНЫМ СВИЩОМ

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Введение. Язвенный колит (ЯК) – это многофакторное хроническое воспалительное заболевание кишечника, характеризующееся такими симптомами, как боль в животе, диарея и ректальное кровотечение. Одним из осложнений ЯК является формирование свищей с мочевым пузырем, влагалищем, промежностью и другими структурами.

Цель исследования – демонстрация клинического случая язвенного колита, осложненного формированием кишечно-кожного свища.

Материал и методы. Данные клинического наблюдения 68-летнего мужчины, находящегося на лечении в отделении гнойной хирургии УЗ «Гродненская университетская клиника».

Результаты. Пациент с низким индексом массы тела (17,1 кг/м²) был госпитализирован с жалобами на выделение кишечного отделяемого из послеоперационного рубца (в анамнезе срединная лапаротомия по поводу кишечной непроходимости с выведением петлевой илеостомы). При проведении фистулографии и колоноскопии выявлены свищевое отверстие в сигмовидной кишке и эрозивный проктосигмоидит. Была выполнена левосторонняя гемиколэктомия. Результаты гистологического обследования соответствовали язвенному колиту. В послеоперационном периоде сформировался абсцесс брюшной полости, который был успешно вылечен дренированием.

Заключение. Случай иллюстрирует сложность лечения язвенного колита и его многофакторную природу.

Ключевые слова*: язвенный колит, воспалительные заболевания кишечника, кишечно-кожный свищ*

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Introduction

Ulcerative colitis, (UC) is a chronic relapsing inflammatory bowel disease (IBD), manifesting various symptoms such as diarrhea, abdominal pain, rectal bleeding with the presence of ulceration of the superficial mucosa. The classical variant of UC is generally manifested by continuous inflammation of the colon starting from rectum and going retrogradely along the colon. Based on the anatomic extend of the inflammation UC can be categorized into pancolitis, left sided colitis or proctitis [1].

Numerous factors may contribute to the development of ulcerative colitis; however, despite decades of research, the exact etiology remains unknown. Additionally, there are still other factors that make it more difficult to identify the underlying causes of ulcerative colitis. This brings the question, if ulcerative colitis might be either a single illness or a group of related or comparable illnesses. Mucosal inflammation in ulcerative colitis patients begins in the rectum and can spread continually to proximal colon segments [2].

This condition is relatively common, affecting individuals across different demographics. Research indicates either an equal distribution between sexes or a slight predominance of females among UC patients. In a study involving 180 cases, women accounted for 55% while men represented 45% [3]. Notably, inflammation in UC is confined to the mucosal layer of the colon [4]. However, in certain cases, such as that of our patient, the inflammation extends through all layers and is accompanied by micro abscesses along the length of the colon.

Objective. To present a clinical case of complicated UC.

Material and Methods

Clinical observation data of a 68-year-old man having been treated in the Purulent Surgery Department of the Grodno University Clinic. He was admitted to the surgical unit because of intestinal discharge from the postoperative scar (with a history of a midline laparotomy for intestinal obstruction with creation of a loop ileostomy).

Results and discussion

We report a case of a 68-year-old male patient with a body mass index (BMI) of 17.1 kg/m2 also having a complex medical history that includes coronary artery disease (CAD), pneumonia, tuberculosis, pulmonary emphysema, and an abdominal aortic aneurysm. This patient was admitted to the hospital when he noticed there had been a leakage of intestinal secretions from the lower edge of a previous laparotomy scar. This surgical intervention had been performed following a prior event of intestinal obstruction and during which a loop ileostomy was established in early 2024, based on an initial diagnosis of nonspecific UC. The opening of the fistula was measured at 0.7cm in diameter. With the primary complaint of intestinal leak, the patient reported general weakness and persistent fatigue which raised concerns regarding his overall clinical status.

A fistulogram was conducted to assess the fistula tract where 30 ml of water-soluble contrast iohexol was injected under X-ray guidance. This imaging confirmed that there was a connection between the defect in postoperative scar with sigmoid colon, providing critical data for subsequent management (Figure 1). A colonoscopy was performed and it revealed erosive proctosigmoiditis and a fistula of the sigmoid colon. A brush biopsy was taken during the procedure which demonstrated moderate leukocyte infiltrate within the colonic mucosa but, it did not show evidence of ulcers or erosions, indicating an inflammatory process without extensive tissue destruction at that time.

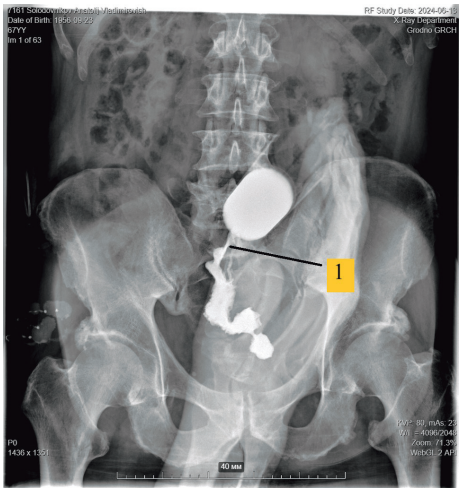

Figure 1. – *Fistulography*
Notation – 1 – colocutaneous fistula.
Рисунок 1 *– Фистулография*
Примечание – 1 – кишечно-кожный свищ.

Further imaging studies were warranted due to the complexity of the findings. An MRI identified free fluid in the right pleural cavity, a simple renal cyst, and a subcapsular hematoma in the liver. A subsequent CT scan provided more thorough information revealing significant changes in the sigmoid colon and adjacent adipose tissue indicative of destructive inflammatory process and identified a structure resembling a fistula. To further support these findings an irrigoscopy was performed which confirmed that the contrast barium did fill the rectum and subsequently exited via the anterior abdominal wall through the identified fistula.

Due to the patient's deteriorating and complex condition, a left sided hemicolectomy, viscerolysis, mesorectumectomy, and end colostomy were performed. Histopathological examination of the biopsy of the left flank of the colon revealed the mucosa with a violation of the architectonics of crypts, the cell population was represented by

diffusely located plasma cells, macrophages, single eosinophils, neutrophils, lymphocytes with the formation of a lymphoid follicle with hyperplasia at the border of the mucous membrane and submucosa. Surface epithelium was with erosion (Figure 2). Histopathology of rectum showed the mucosa of the colon with the formation of mucin granuloma, represented by fragments of destroyed epithelial cells and necrotic tissue, mucin with an abundance of segmented neutrophils. The granuloma was surrounded by a shaft of abundant lymphocytes and scattered eosinophils (Figure 3). Pericolic tissues were with granulomatous reaction - the outcome of ulceration with the formation of deep cracks. Fatty tissue was with polymorphocellular inflammatory infiltration and the presence of fatty necrosis with the formation of foreign body granules. There was a large number of xanthoma cells with foamy cytoplasm containing resorbed fatty inclusions (Figure 4). So, in all samples widespread granulomatous inflammation along with features suggestive of ulcerative colitis was revealed. Postoperatively, laboratory results showed that C reactive protein (CRP) was elevated signaling a severe inflammatory response and mild anemia was also noted (Table 1).

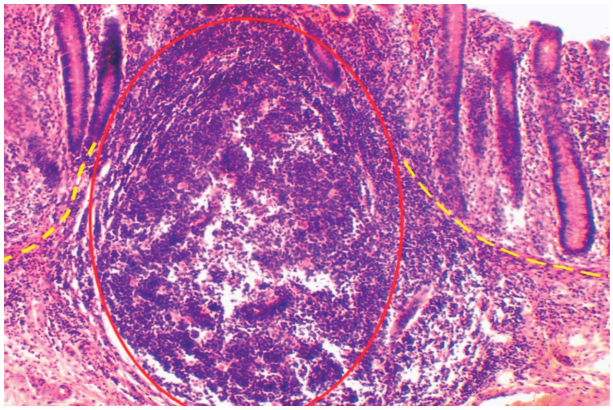

***Figure 2** – Histopathology of the left flank of the colon. Staining: hematoxylin and eosin. ×40.*
Notation: red oval - lymphoid follicle with hyperplasia, yellow dotted lines - the border of the mucous membrane and submucosa.
***Рисунок 2** – Гистология левого фланга ободочной кишки. Окр.: гематоксилином и эозином. ×40*
Примечание – красный овал – лимфоидный фолликул с гиперплазией, желтые пунктирные линии – граница слизистой и подслизистой оболочки.

The APACHE II score was taken into consideration of the patient and it was recorded at 12, indicating a 14.6% risk of mortality. The SOFA score was recorded at five reflecting multi organ dysfunction. In order to assess the kidney function, the CKD-EPI formula was used to calculate the estimated glomerular filtration rate (eGFR) and it was 48ml/min/1.73 $m^{2.}$ It indicted that there was a decrement of its function. Additionally, post operative procalcitonin levels were elevated at 12.8ng/ml, indicating significant systemic infection or inflammation (Table 1).

As a response to the patient's deteriorating conditions characterized by severe intoxication,

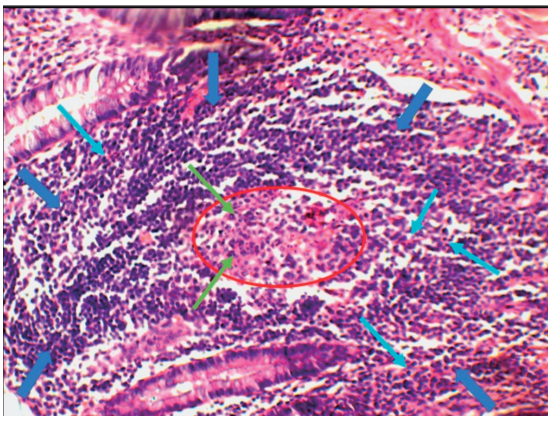

***Figure 3** – Histopathology of rectum. Staining: hematoxylin and eosin. ×100.*
Notation: red oval – mucin granuloma, green arrows - mucin with an abundance of segmented neutrophils, blue arrows – lymphocytes, light blue arrows - eosinofils
***Рисунок 3** – Гистология прямой кишки. Окр.: гематоксилином и эозином. ×100*
Примечание – красный овал – муциновая гранулема, зеленые стрелки – муцин с обилием сегментоядерных нейтрофилов, синие стрелки – лимфоциты, голубые стрелки – эозинофилы

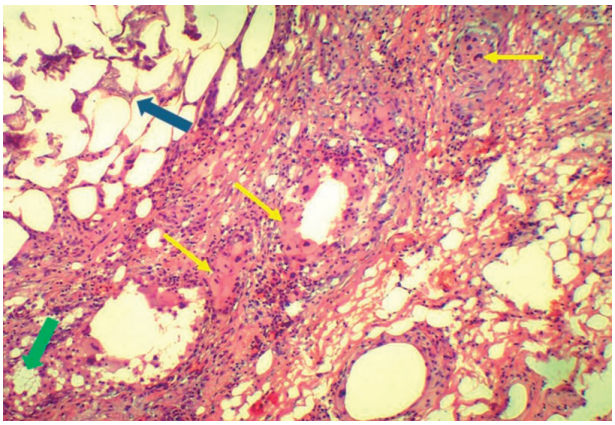

***Figure 4** – Histopathology of pericolic tissues. Staining: hematoxylin and eosin. ×100.*
Notation: blue arrow - fatty necrosis, yellow arrows - xanthoma cells with foamy cytoplasm, green arrow - resorbed fatty inclusions.
***Рисунок 4** – Гистология периколической клетчатки. Окр.: гематоксилином и эозином. ×100*
Примечание – синяя стрелка – жировые некрозы, желтые стрелки – ксантомные клетки с пенистой цитоплазмой, зеленая стрелка – резорбированные жировые включения.

hemadsorption was performed utilizing the sorbent Hemoproteasorb. A total volume of six liters of heparinized blood (1000 IU) was used and this procedure was repeated several times due to the critical condition of the patient.

Postoperative period was complicated by hyperthermia, raising concerns for infections. A CT scan which followed revealed an abdominal abscess. An ultrasound guided incision and drainage procedure was performed under local anesthesia where thick foul-smelling pus was successfully evacuated. To ensure adequate drainage from the abscess cavity a PVC tube was placed. A microbiological examination of a smear taken from the puncture of the abscess cavity

Table 1 – Laboratory findings during hospitalization
Таблица 1 – Лабораторные показатели во время госпитализации

Parameter	On the day of admission	3rd Day after operation	1 month after operation	Reference values
RBC, x10^{12}/L	5,51	2,63	3,0	3,9-5,1
Hct, %	47,7	24	26	35-50
Hb, g/L	151	76	81	130-170
WBC, x10^{9}/L	6,46	6,2	4,46	4-9
PLT, x10^{9}/L	322	140	379	150-450
CRP, mg/L	16,8	394,7	38,8	0-6
Amylase, U/L	49	42	-	25-100
Total protein, g/L	73	50	59	65-85
Fibrinogen, g/L	4,79	8,07	-	2,7-4,7
Creatine, μmol/L	60	94	64	62-124
APTT, sec	25,3	53,8	-	22-35
PCT, ng/mL	-	>12,8	0,105	0,5
D dimer, ng/ml	-	2,28	-	0,063-0,701

Notation: RBC- Red blood cells, Hct - Hematocrit, Hb - Hemoglobin, WBC - White blood cells, CRP - C Reactive Protein, APTT - Activated Partial Thromboplastin Time, PCT – Procalcitonin.

Примечание – RBC – эритроциты, Hct – гематокрит, Hb – гемоглобин, WBC – лейкоциты, PLT – тромбоциты, CRP – С-реактивный белок, APTT – активированное частичное тромбопластиновое время, ПКТ – прокальцитонин.

identified the microorganism Proteus mirabilis (10^6). An antibiogram was performed in order to navigate the appropriate treatment regime. It revealed that this particular strain was resistant to many antibiotics' combinations. These included Ceftriaxone, Cefepime, amoxicillin-clavulanic acid, Co-trimoxazole, meropenem, nitrofurantoin, Ciprofloxacin, tigecycline and ampicillin. The strain was sensitive for amikacin and gentamicin and leading the way for effective management.

The patient was stabilized following the above interventions. Vitals were pulse 82/min, and respiratory rate 18/min and 125/80 mmHg, without signs of peritoneal irritation. Induction therapy was initiated with mesalazine at a dosage of 3g/day orally for his inflammatory bowel, alongside Tardyferon 80mg daily before meals for three months for his anemia. Upon the patient's clinical improvement and stabilization, he was discharged with instructions regarding medication compliance, dietary modifications and the necessity for regular follow up appointments to monitor his recovery and manage any potential complications (Figure 5).

The final diagnosis was "Nonspecific Ulcerative colitis with Formation of Paracolytic Infiltrate. Colon obstruction. Loop ileostomy from 19.01.2024. Erosive proctosigmoiditis. Laparotomy, viscerolysis, resection of a portion of descending colon, sigmoid colon with fistula, mesorectumectomy, descendostomy 05.07.2024. Abscess of the abdominal cavity. Abscess drainage 24.07.2024. Sepsis."

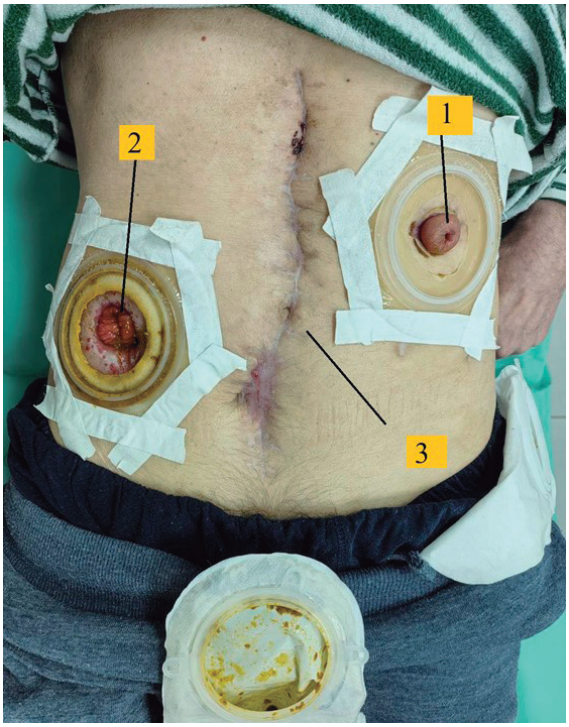

Figure 5 *– Patient at the time of discharge. Notation: 1 – colostomy, 2 – ileostomy, 3 – postoperative scar.*
Рисунок 5 *– Пациент в день выписки из стационара. Примечание: 1 – колостома, 2 – илеостома, 3 – послеоперационный рубец.*

Fistulous communication with surrounding structures such as bladder, vagina, perineum and ischiorectal fossa are complications that can arise from inflammatory colonic pathologies [5]. Surgical intervention, in lower gastrointestinal tract involving

anastomosis, suturing and stapling carries a high risk for acute and chronic leaks, particularly following procedures for inflammatory bowel disease or irradiated colorectal cancer [6]. Colocutaneous fistulas are communications between skin of abdominal wall and the colon. Around 10-30% of gastrointestinal fistulas are colocutaneous fistulas, these can be further classified as postoperative or spontaneous based on the etiology of their development. Approximately 25% of spontaneous fistula usually develops often in association with cancer, radiation therapy or inflammatory conditions such as diverticular disease, bowel diseases due to ischemia, IBD, with IBD being the most frequent cause. The remaining 75% arises postoperatively, following surgeries for adhesive bowel obstruction, IBD or malignancy. Healing of these fistulas are hampered due to factors such as high output, significant disruption of intestinal continuity and active IBD in affected segments [7]. Moreover, delayed wound healing of fistulas depends on individual dependent factors such as diabetes mellitus, smoking obesity and anatomical complexities surrounding type fistulas and post operative infections [8].

Like in the case of our patient who presented with proctosigmoiditis a colocutaneous fistula developed communicating with the sigmoid colon. Moreover, correlation studies have shown a relationship between age and the coexistence of CAD and IBD. One investigation comprising of 524 matched non-IBD controls with CAD and 111 IBD patients put of which 54 had Crohn's disease and 77 suffered from UC, found that IBD patients were younger (mean age 65.3 +/- 11.0 years) compared to CAD alone (67.8+/- 11.0 years, P=0.016), suggesting an association between CAD and IBD in a younger demographic [9].

Diverticular colitis is a condition where there is chronic inflammation of colon in the presence of diverticula. There had been several studies that have documented cases where diverticular colitis has been progressively manifested into UC both endoscopically and histologically [10].

Given this, in our patient the discovery of a diverticula raised the possibility of diverticulitis contributing to the onset of UC and /or fistula formation; however, there was insufficient evidence to substantiate this hypothesis.

Furthermore, a recent case series of patients with septic shock and renal failure receiving hemadsorption treatment showed a positive hemodynamic stabilization following the hemadsorption treatment, as well as the fact that the stabilization was rapid and increased the survival [11]. This was as well demonstrated by our patient who had a low GFR and was admitted to the intensive care unit after developing sepsis postoperative to the left -sided hemicolectomy. The patient's APACHE II score valuing a score of 12 indicated high mortality risk [12]. The type of hemosorbent used in this instance was Hemoproteazosorb, which was chosen because of its effective anticytokine qualities as demonstrated by research conducted by members of our medical institute [13, 14].

Moreover, during his septic condition our patient was seen to have a marked elevation of procalcitonin level of 12.8ng/ml (< 0.1 ng/ml) postoperatively, which supports the idea of studies that show PCT as a promising marker of sepsis, able to supplement clinical signs and routine lab parameters suggesting severe infection [15].

Conclusions

This case report shows the complexities in management of UC and its complications one of which is the development of colocutaneous fistula. Our patient's complex medical history, severity of UC, surgical intervention and associated complication such as abscess formation and sepsis signify the importance of due diligent treatment tactics.

Furthermore, the association of IBD and CAD calls for the need for vigilance in monitoring of patient with UC for comorbidities.

The successful management of our patient using Hemoproteazosorb as a hemadsorption therapy demonstrates the potential for innovative strategies on improving clinical outcomes in patients with sepsis. Moreover, timely treatment of diverticula and UC at an early stage improves significantly the outcome of bowel health and patient's quality of life.

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Conflict of interest. The authors declare no conflict of interest.

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Conformity with the principles of ethics. The study was approved by the local ethics committee. Written informed voluntary consent was obtained from the patient for the publication of the case report and publication of photographic materials in a medical journal, including its electronic version (date signed by patient – 02.10.2024).

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