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CASE REPORT / ПРИКАЗ БОЛЕСНИКА

Solitary cecal ulcer – case report and treatment options according to literature review

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SUMMARY

Introduction Solitary cecal ulcer is a benign and extremely rare disease, as less than 300 cases have been reported so far. The etiology is unknown, and it can be diagnosed by a pathohistological examination. Often presented as an acute abdomen and rectal bleeding, it can mock various important and urgent conditions. Treatment protocol is not defined. Extensive and radical surgeries are often performed due to this benign disease mimic. Our aim was to indicate this disease, present the treatment, and to facilitate the treatment plan for the disease.

Case outline A 67-year-old female patient was admitted to the Emergency Department with a clinical manifestation of acute appendicitis. Emergency surgery was indicated by the diagnostic tests. The intraoperative finding revealed an ulcer on the cecum, which was sutured. The patient fully recovered, and subsequent colonoscopy and pathohistological findings indicated a solitary ulcer.

Conclusion It is possible to treat this condition by retaining the organ and avoiding major surgery. Therefore, it is our opinion that it might provide significant assistance to clinicians in a similar situation. Hence, it is undoubtedly an interesting case for archiving, especially since such a case had not been recorded in our country previously.

Keywords: ulcer; rare disease; cecum; colonic disease

INTRODUCTION

Solitary cecal ulcer (SCU) is a rare, benign, and specific entity. It is described only as a case report or, in some rare cases as case series. SCU is the ulcer of the cecum without common etiology following pathohistological examination. Between 250 and 350 cases of solitary colonic ulcers have been reported in the world thus far, whereas just over 258 cases have been detected in the cecum. It was noted in subject-specific medical publications that about 67% of this disease affects the cecum, 18% transverse colon, hepatic, and splenic flexure, and 15% descending and sigmoid colon [1].

Cruveilhier described SCU for the first time in 1832 [2, 3]. It is a rare disease and it can easily be superseded by acute appendicitis or colonic neoplasm. In most cases, it can involve conservative treatment rather than surgical, except in cases of perforation, obstruction, and uncontrolled bleeding. The dominant symptom is a pain in the lower right quadrant of the abdomen. It is diagnosed as an acute appendicitis in 50% of cases. It can be identified as lower gastrointestinal hemorrhage (33%), visceral perforation (19%), or palpable abdominal mass (16%) [1, 3].

Physical examination in most cases reveals tenderness in the lower right quadrant of the abdomen, and in some cases it might be reflected as an acute abdomen or rectal bleeding. Laboratory tests are nonspecific; inflammation markers can be elevated, blood count may be lowered, tumor markers are not elevated. Radiographic imaging is usually nonspecific or it can show bowel obstruction or pneumoperitoneum due to perforation. Ultrasound of the abdomen is also nonspecific or it can show a mass in the cecal region. Computed tomography usually shows wall thickening of the cecum. The best way to diagnose SCU is by conducting colonoscopy screening, followed by a pathohistological examination of a biopted or resected sample. Findings are usually nonspecific chronic inflammation, and rarely an acute inflammation. There is no substantial reasoning about etiology in these samples. It is speculated that long-term use of non-steroidal anti-inflammatory drugs (NSAID) is the main causative agent of SCU.

The typical position of the ulcer is lateral, anti-mesenteric, on the cecum wall, 2 cm cranial from the ileocecal valve [4, 5, 6].

For this study, it was of interest to point out this disease, to present how we have managed this rare condition, and to contribute to defining a treatment protocol for this disease.

CASE REPORT

A 67-year-old female patient was admitted to the Emergency Department with the main complaint of severe pain in the right lower quadrant of the abdomen. The pain started two weeks before the admission, when the pain became unbearable. She experienced nausea

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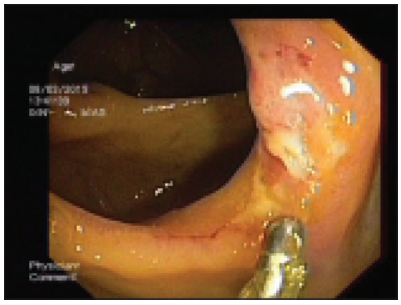

Figure 1. Colonoscopy finding before biopsy

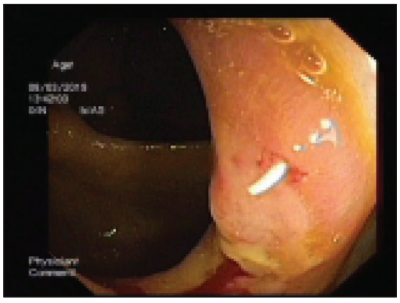

Figure 2. Colonoscopy finding after biopsy

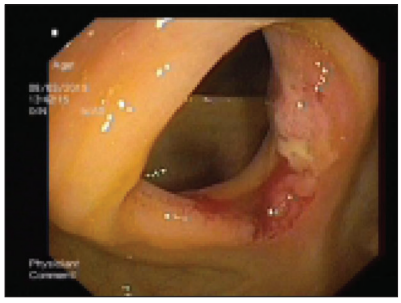

Figure 3. Colonoscopy finding after biopsy

and vomiting. Rectal bleeding, weight loss, and other complaints were not detected. The patient reported several comorbidities – hypertension, ischemic cardiomyopathy, diabetes mellitus, and previous myocardial infarction. The patient regularly used the following medicines: isosorbide mononitrate, trimetazidine, a combination of clopidogrel and acetylsalicylic acid, the combination of ramipril and felodipine, bisoprolol, atorvastatin, metformin, and gliclazide. She neither smoked for the last eight years nor used alcohol.

The patient's health status, on admission, was good – she was normotensive, afebrile, with normal heart rate and blood oxygen saturation. During palpation, we discovered tenderness in the lower right quadrant of the abdomen and rebound pain, with peritoneal irritation and without abdominal rigidity. There were no other signs of the aforementioned condition, and the rest of the clinical findings were satisfactory. Laboratory tests showed an elevated number of white blood cells, 19.3×10^9/L with predomination of granulocytes 85% or 16.6×10^9/L, elevated glycemia 9.32 mmol/L, and C-reactive protein 106 mg/L; on the other hand, hemoglobin and hematocrit were lowered: 114 g/L and 34.4%, respectively. There were no pathological changes in the urine. Ultrasonography of the abdomen and urinary system showed cholecystolithiasis (without inflammation), meteorism, dilatation of the right pyelocalyx (grade I), without other pathological changes; however, the appendix could not be displayed. Abdominal radiography showed meteorism with hydroaeric level in the right iliac fossa. The emergency surgery was indicated due to suspected acute appendicitis with the risk of perforation.

Intraoperatively, a small amount of turbid whitish fluid was found, however, the swab test was sterile. There was an ischemic field on the lateral cecal wall, with an approximate diameter of 35 mm. Those areas were thin, dark, and deserosed with signs of local peritonitis and reactive appendicitis. We stitched that field in two layers with polydioxanone 3.0 suture in a continued and interrupted manner. Following appendectomy, flushing and drainage were applied. During the postoperative period, the patient was hemodynamically stable, afebrile, in good overall condition with satisfactory local findings. Laboratory tests showed a decline in inflammatory markers. The patient was discharged from the hospital after seven days, receiving a recommendation for further therapy and a colonoscopy screening in two months. The overall condition of the patient was satisfying during subsequent check-ups. Three weeks after the surgery, the results of pathohistological analysis of the appendix showed fibrous obliteration of the appendix lumen. Three months after surgery, the pathohistological analysis of findings of suspected biopted change observed during the colonoscopy showed a separation of the wide and shallow lesion on the fold in front of the valve, resembling an ulcer covered with fibrin; the rest of the colon and rectum were free of pathological changes (Figures 1, 2, and 3).

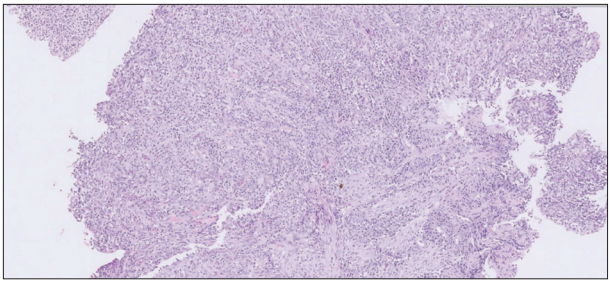

Figure 4. The bottom of the ulcer (H&E, × 10)

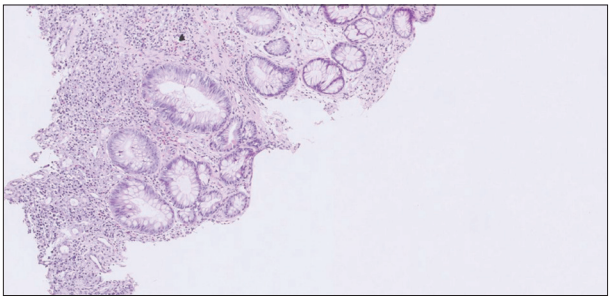

Figure 5. Colonic mucosa with dystrophic crypts and laminae propriae fibrosis in the lower part of the image (H&E, × 10)

The pathohistological finding of Prof. Slavica Ušaj, M.D. (pathologist, subspecialist cytologist) was as follows: the samples consist of fragments of necrotic detritus and fragments of colonic mucosa with ulceration of the entire thickness of the mucosa; the bottom of the ulcer makes nonspecific granulation tissue imbued with a mixed inflammatory infiltrate; in the surrounding mucosa, crypts are distorted and lined with regenerative epithelium, elongated and pseudo-stratified nuclei; one focus loses maturation to the surface and the same type of change is found in a small portion of the superficial epithelium. Therefore, she concluded that it was a solitary cecal ulcer with the focus of low-grade dysplasia (Figures 4 and 5).

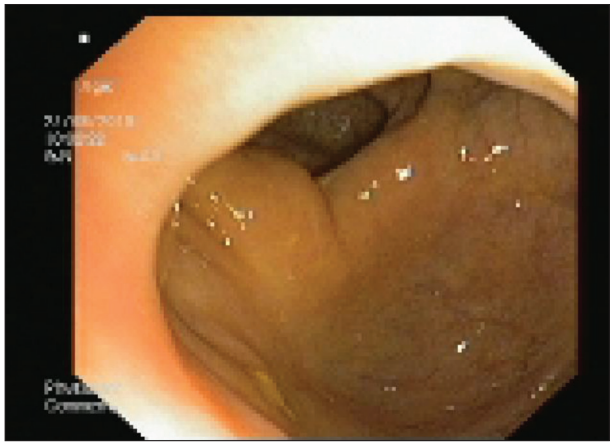

Figure 6. Control colonoscopy finding

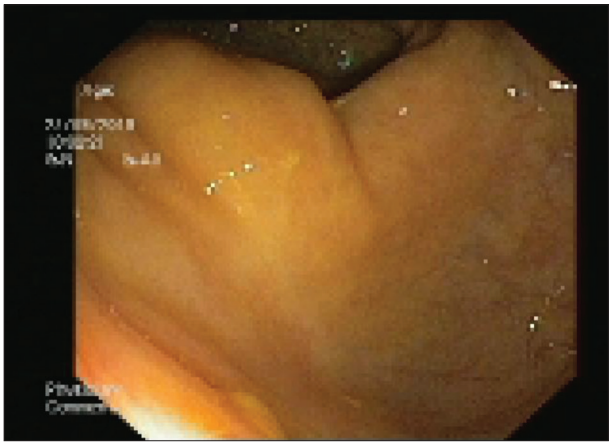

Figure 7. Control colonoscopy finding

Six months after the surgery, the patient did not experience any pain. She went for a control colonoscopy screening nine months after the surgery. The findings showed complete healing of the ulcer, the mucosa had normal appearance; laboratory findings were within the reference ranges, except for the blood count, which was slightly lower than the lower limit of the reference values (Figures 6 and 7).

For the publication of this case, we received written consent of the patient and the Hospital Ethics Committee No 01-797/9.

DISCUSSION

Referencing the available literature, we discovered a total of 258 reported cases of solitary cecal ulcer. We searched through PubMed database (148 cases reported in case reports and case series), Google Scholar database (110 cases reported in case reports and case series), Cochrane database (there were no systematic reviews and meta-analyses about solitary cecal ulcer), and Scopus database (no reports). We were looking for cases of a cecal ulcer and a solitary cecal (coecal) ulcer (ulcus) in these databases and in the references of the publications that we found. We excluded transplanted patients due to an altered immune system, in which the cause of the ulcer is usually cytomegalovirus (CMV) [5]. Transplanted and dialysis patients have a high mortality rate (50%) if surgery is required [7].

Our patient was a 67-year-old female; analysis of other studies and their findings indicate that the sex ratio is usually 50:50 [8, 9]. The median age that we found in the case series and reports is 57 years, which is similar to the results of some studies that represented age predilection of 40–60 years [1] or age median of 61 years [8].

The clinical appearance and laboratory findings of our patient suggested that she suffered from acute appendicitis. Emergency surgery was indicated. Colonoscopy or resection of the specimen is necessary for pathohistological confirmation, which is the only method to confirm this entity. In our case, it was not indicated because of the clinical picture of the acute abdomen and the risk of potential complications it brings along, as well as because of the complexity of delivery in unprepared patients. In any other case – rectal bleeding, suspected tumor, pain of unclear etiology – colonoscopy is crucial [3].

We opted for laparotomy instead of laparoscopy due to suspected perforation and purulent peritonitis. We were surprised by the findings since the change on the cecum wall was not clear to us, and our first thought was ischemia due to micro-embolization or the action of some aggressive agent. The limitation of change was clear and proper. There was no perforation.

According to some authors, a possible etiology of SCU is limited ischemia, caused by vasculitis and/or microembolization of the terminal branches of the colon nutritional arteries [9, 10]. The most frequently mentioned cause of SCU are NSAID drugs [11], but we found only a few cases of SCU in patients on NSAID therapy. For other patients, it is either unknown whether they used NSAID or not. Our patient also did not use NSAIDs, but she used acetylsalicylic acid for several years.

We decided to preserve the colon and provide the lesion with seromuscular sutures in two layers. The integrity of the wall, the vital edges of the ulcer, and the absence of a palpable tumor were reasons to think that suturing the ulcer and preserving the colon was a good solution, and perhaps the best one. The appendectomy was executed due to inflammatory altered walls. It was most likely a consequence of regional inflammation. Two authors performed similar surgery in cases of SCU with the clinical findings of appendicitis or perforation [12, 13]. In the earlier period, surgical treatment of this entity was insisted on [14], while in recent times, conservative treatment has been favored, except in cases of perforation, uncontrolled bleeding, or acute abdomen [4, 15]. The range of applied operations is wide. The most common is right hemicolectomy, open or laparoscopic, about 41% of all operations duo to SCU according to the available literature [16]. Other operations – segmental resection and stoma, ileocecectomy and anastomosis, cecostomy due to perforation, laparoscopic or open sleeve cecectomy, open or laparoscopic-assisted ulcer excision, or even total colectomy – are rarely performed. Conservative therapy includes symptomatic therapy, blood replacement if necessary, and regular colonoscope monitoring, but without exactly specified intervals. Two authors

presented two successful treatments with antibiotics, ciprofloxacin, and metronidazole [4, 9].

A big problem is pathohistological confirmation, as it is the only way to confirm SCU with certainty. The acute condition represents an even larger issue. In most such cases, pathohistological confirmation is not possible. Also, there is a growing possibility that a larger and more radical operation will be performed due to benign disease. It is very difficult to prevent such an outcome. According to the experience of several authors so far, in cases of accidental discovery, pathohistological verification and conservative treatment is the best option. But the question what to do in case of an acute condition remains.

Our case confirmed that minimal surgical intervention with organ preservation is possible. The organ and its function can be preserved completely, facilitating a better quality of life for the patient. A significant benefit is that there is less chance of complications which happen after major resection procedures, such as non-healing of the anastomosis, enteral fistula, peritoneal cavity infection, wound infection, septicemia, multi-organ failure, and death. Lesser invasiveness and lower number and extent of complications decrease the treatment expenses, duration of hospitalization, increase the odds for positive treatment outcome and shorten the period of recovery, thus enabling an earlier return to regular daily activities.

Colonoscopy monitoring and pathohistological verification is mandatory after the intervention. It remains to determine time intervals. The main dilemma remains as to when malignancy is suspected in acute conditions – whether it is justified to take a clip and do an extempore biopsy, or should we adhere to oncological principles. On the one hand, it is a quite rare entity, and the possibility of malignity is very high; on the other hand, what matters most is each patient's life and its quality.

Conflict of interest: None declared.

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Солитарни улкус цекума – приказ случаја и терапијске могућности према прегледу литературе

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САЖЕТАК

Увод Солитарни улкус цекума је бенигно и екстремно ретко обољење; до сада је пријављено мање од 300 случаја. Узрочник обољења је непознат, а може бити потврђено само патохистолошким испитивањем. Често је презентовано као акутни абдомен и ректално крварење, а може имитирати и разна друга битна и ургентна стања. Не постоје протоколи лечења за ово обољење, а често се изводе велике и радикалне операције због њега.

Наш циљ је да представимо како смо решили један такав случај.

Приказ болесника Жена старости 67 година примљена је у Ургентни центар са сликом акутног апендицитиса. Након урађене дијагностике индикована је хитна операција. Интраоперативни налаз је указивао на улкус цекума, који смо прешили, чиме смо сачували орган. Болесница се у потпуности опоравила, а каснији колоноскопски и патохистолошки налаз је показао да је у питању солитарни улкус цекума.

Закључак Могуће је сачувати орган и избећи већу операцију код постојања овог обољења. Мишљења смо да би овај случај могао помоћи клиничарима који се нађу у сличној ситуацији. Свакако, случај је занимљив за архивирање, поготово што у нашој земљи још није забележен овакав случај.

Кључне речи: улкус; ретко обољење; цекум; обољење колона