

A Case of Ulcerative Colitis Complicated by a Simple Appendiceal Opening

Xiao Liu¹, Fang Fang¹, and Qingfeng Luo¹

¹Beijing Hospital

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Xiao Liu^{1*}, Fang Fang², Qingfeng Luo¹

¹ Department of Gastroenterology, Beijing Hospital, National Center of Gerontology; Institute of Geriatric Medicine, Chinese Academy of Medical Sciences, P.R. China

² Department of pathology, Beijing Hospital, National Center of Gerontology; Institute of Geriatric Medicine, Chinese Academy of Medical Sciences, P.R. China

Email: *liuxiao3424@bjhmoh.cn*

Abstract

This case report describes the clinical course of a 64-year-old male with intermittent abdominal pain attributed to recurrent ulcers at the appendiceal orifice. Initial investigations in November 2019 revealed chronic gastritis and ulcers at the appendiceal orifice, prompting consideration of ulcerative colitis. The patient responded well to mesalazine therapy, experiencing relief from symptoms and improved colonoscopy findings in May 2020. Despite discontinuing medication, a recurrence of symptoms in August 2021 led to a repeat colonoscopy showing renewed ulcers. Mesalazine was reinstated, resulting in symptom resolution and improved colonoscopy findings by December 2021. However, in May 2023, a subsequent recurrence of abdominal pain and colonoscopy-confirmed mucosal changes at the appendiceal orifice prompted reintroduction of mesalazine. The patient remains under regular monitoring on mesalazine therapy. This case highlights the challenges in managing recurrent appendiceal ulcers and the importance of long-term therapeutic vigilance in suspected ulcerative colitis cases.

Keywords ulcerative colitis, appendiceal orifice, mesalazine

INTRODUCTION

Ulcerative colitis (UC) is a chronic non-specific inflammatory disease of the gastrointestinal tract, often originating in the rectum and progressing retrograde to involve the entire colon and the terminal ileum. Affection of the appendix opening is observed in some UC patients, although isolated involvement of the appendix in UC is rarely reported. Herein, we present a case of recurrent UC with exclusive involvement of the appendix, highlighting that clinical manifestations of UC may atypically manifest as isolated appendiceal involvement. Isolated inflammation of the appendix mucosa is a characteristic feature of ulcerative colitis and may serve as an indicator of disease activity and prognosis. This case report examines the intricate clinical journey of a 64-year-old male with intermittent abdominal pain attributed to recurrent ulcers localized at the appendiceal orifice. The patient's diagnostic odyssey began with findings of chronic gastritis and subsequent identification of appendiceal ulcers, prompting suspicion of ulcerative colitis. The report tracks the patient's response to mesalazine therapy, the recurrence of symptoms despite initial improvement, and the therapeutic

challenges encountered in managing this unique manifestation. This case underscores the complexities in diagnosing and managing ulcerative colitis, especially when marked by recurrent focal lesions, emphasizing the significance of continual vigilance and individualized therapeutic approaches.

CASE REPORT

A 64-year-old male presented with intermittent abdominal pain. He had a history of gastroesophageal reflux disease (GERD) but no other significant medical history. In November 2019, the patient initially sought medical attention due to intermittent right lower abdominal pain and sporadic loose stools. Subsequent investigations revealed chronic gastritis upon further evaluation with esophagogastroduodenoscopy, and colonoscopy indicated the presence of ulcers at the appendiceal orifice (**Figure 1**). There were no apparent signs of significant erythema, erosion, or ulceration in other areas of the colon. Capsule endoscopy and abdominal-pelvic CT scans showed no notable abnormalities. Biopsy of the ulcerated appendiceal orifice during colonoscopy revealed moderate chronic mucosal inflammation with mild active changes, glandular hyperplasia, lymphoid tissue hyperplasia, and evidence of acute cryptitis (**Figure 2**).

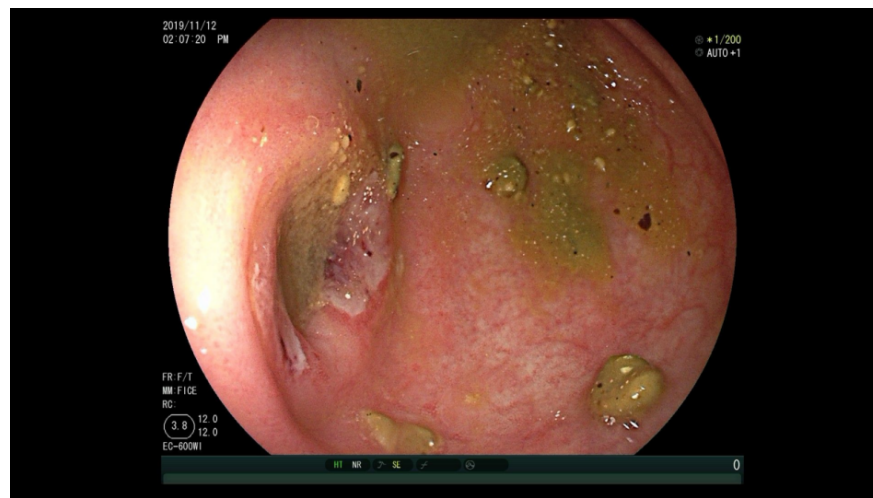


Figure 1 Colonoscopy in November 2019: Appendiceal orifice

A B

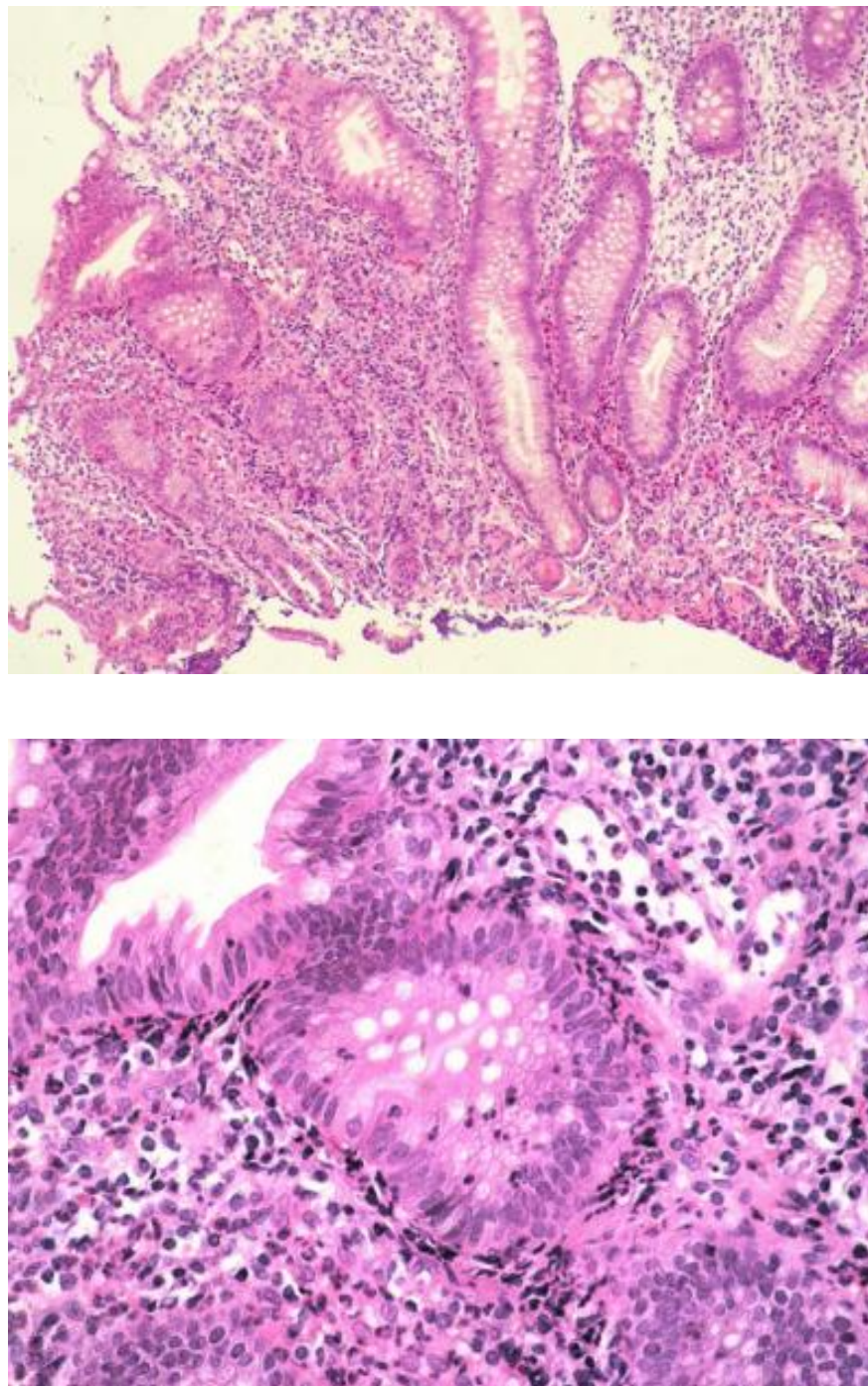


Figure 2 . HE staining showed moderate acute and chronic inflammation in the appendiceal orifice mucosa, accompanied by acute cryptitis. A, 100x magnification; B, 200x magnification.

Considering the patient's clinical history and endoscopic findings, ulcerative colitis was suspected, and the patient was initiated on a daily oral dose of 3g mesalazine. The patient reported gradual relief from abdominal pain, and his bowel movements became regular and formed. A follow-up colonoscopy in May 2020 showed improvement in the appearance of the ulcers at the appendiceal orifice, with scattered areas of congestion

and erosion but no significant ulcer formation (**Figure 3**). Biopsy results indicated severe acute and chronic mucosal inflammation with erosions, interstitial eosinophilic infiltrates, and focal granuloma formation. The patient, feeling significantly improved, discontinued the medication on his own.

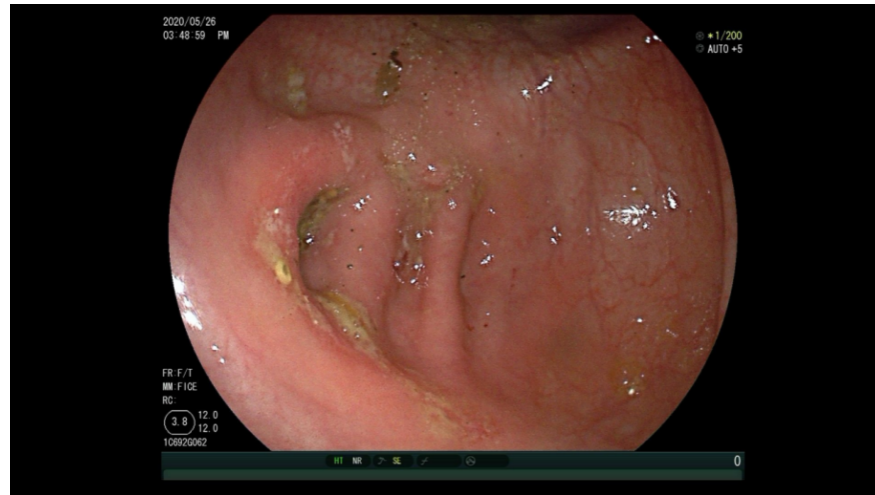


Figure 3. Colonoscopy in May 2020: Appendiceal orifice

However, in August 2021, the patient experienced a recurrence of symptoms, including intermittent right lower abdominal pain, with no fever or changes in bowel habits. A repeat colonoscopy showed a recurrence of ulcers at the appendiceal orifice (**Figure 4**). The patient was once again treated with mesalazine, leading to symptom relief. In December 2021, a follow-up colonoscopy demonstrated a marked improvement in the appearance of the ulcers at the appendiceal orifice (**Figure 5**). The patient reported the disappearance of abdominal pain and continued taking mesalazine regularly for a year.

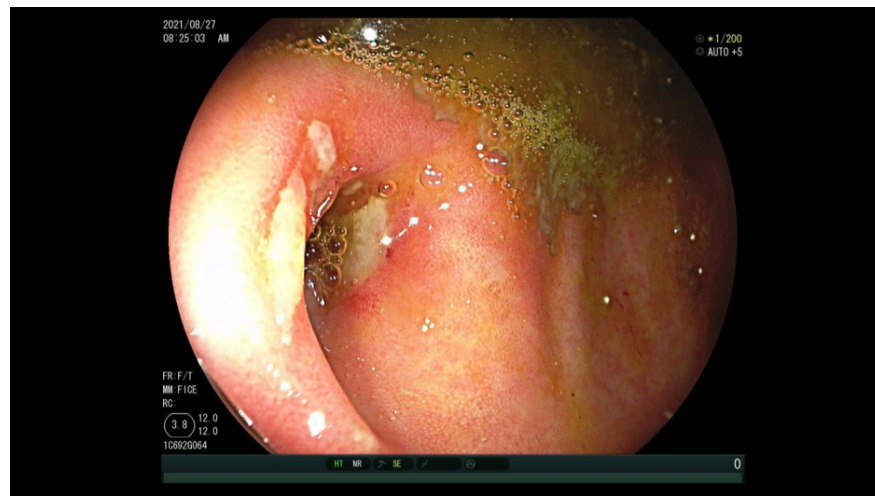


Figure 4. Colonoscopy in August 2021: Appendiceal orifice

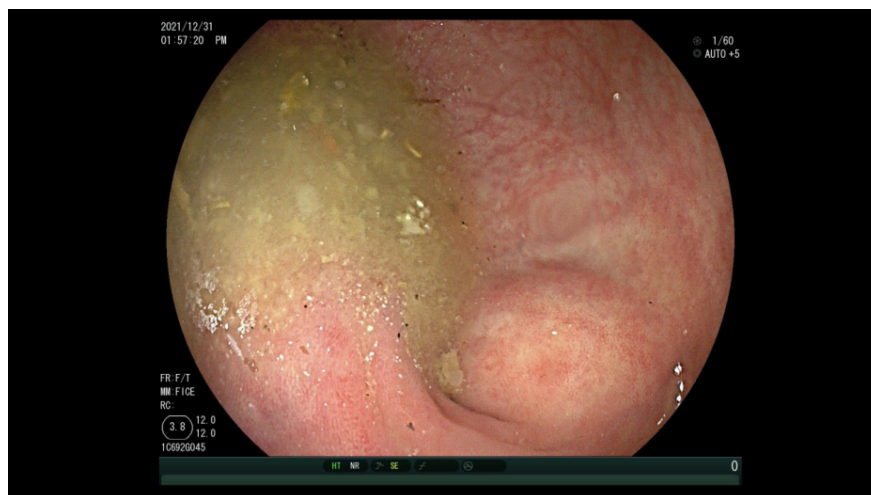


Figure 5 Colonoscopy in December 2021: Appendiceal orifice

Subsequently, the patient experienced recurrent intermittent right lower abdominal pain, with no fever or alterations in bowel habits. In May 2023, a repeat colonoscopy indicated mucosal congestion and erosion at the appendiceal orifice (**Figure 6**). Histopathological examination revealed acute and chronic mucosal inflammation, superficial epithelial changes with erosions, and shallow ulceration, along with evidence of cryptitis and an increased number of lamina propria plasma cells. The mucosal surfaces in other parts of the colon remained unremarkable. Mesalazine was reintroduced, resulting in the resolution of abdominal pain symptoms, and the patient continues to be regularly monitored on mesalazine therapy.

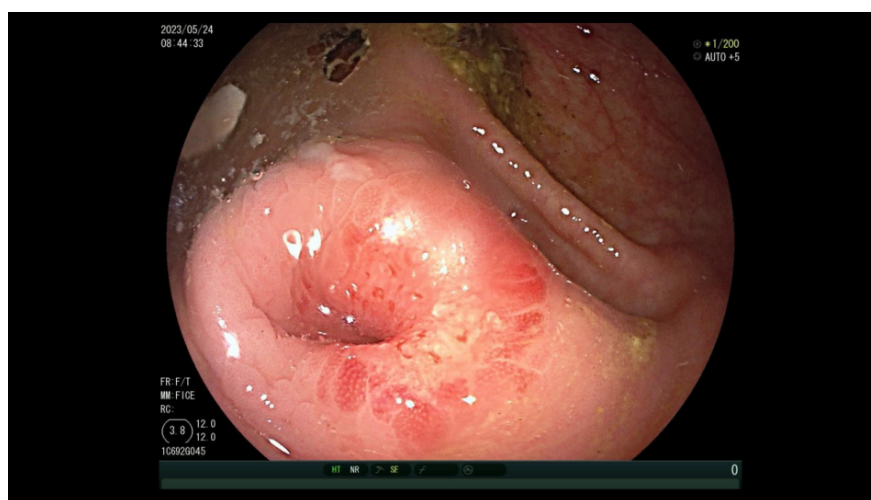


Figure 6 Colonoscopy in May 2023: Appendiceal orifice

3. DISCUSSION

Ulcerative colitis (UC) is a chronic non-specific inflammatory bowel disease characterized by recurrent symptoms such as abdominal pain, diarrhea, and bloody mucous stools. The disease typically follows a chronic course with alternating periods of flare-ups and remission, and in some cases, symptoms may persist and gradually worsen. UC usually begins in the rectum and progresses proximally, potentially affecting the entire colon, including the terminal ileum. Appendiceal orifice inflammation (AOI) refers to congestion, erosion,

and ulcers at the appendiceal orifice. When AOI co-occurs with UC, its inflammatory characteristics resemble the pathological features of UC. There is increasing evidence to suggest that AOI represents a skip lesion in the mucosa of UC [1]. Research indicates that AOI is more common in UC patients with mild to moderate distal colonic involvement [2]. However, retrospective studies have shown conflicting results, with some suggesting that AOI is more prevalent in moderate to severe UC, possibly serving as a marker of active disease and an indicator of disease severity [3].

The presence of AOI in UC patients has been associated with differing outcomes and responses to treatment. Wu and colleagues conducted a retrospective study involving 202 UC patients, of which 116 had AOI. They found a significant difference in the cumulative risk of endoscopic complete remission between the AOI and non-AOI groups, indicating a lower rate of endoscopic complete remission in UC patients with AOI [4]. In a long-term follow-up study by Oh and colleagues involving 318 UC patients, 140 of whom had AOI, the AOI group demonstrated a lower rate of endoscopic complete remission compared to the non-AOI group. However, there were no significant differences between the two groups in terms of other clinical parameters, including the use of biologics, hospitalization rates, and proximal disease extension [5]. Conversely, a retrospective analysis by Kyong and colleagues, involving 376 UC patients with an average follow-up time of 66.1 months, revealed no significant differences between the AOI and non-AOI groups in terms of endoscopic remission, hospitalization rates, recurrence rates, or the use of corticosteroids, immunosuppressants, and biologics [6]. Therefore, the role of AOI in predicting the prognosis of UC patients remains inconclusive.

Pathologically, AOI does not possess specific characteristics. A study analyzing 26 cases with histological abnormalities at the appendiceal orifice found active inflammation in 12 cases, chronic active inflammation in 13 cases, and one case resembling collagenous colitis. Additionally, eight patients with inflammation in other biopsy samples were eventually diagnosed with ulcerative colitis, with none of the patients isolated to the appendiceal orifice showing clinical symptoms during follow-up. As such, isolated inflammation of the appendiceal orifice mucosa should not be regarded as a sign of inflammatory bowel disease progression or as a distinguishing feature of other chronic colonic inflammations [7]. Nevertheless, while AOI is not uncommon in UC patients, it is often reported as a manifestation of "skip inflammation" within UC, and cases of isolated AOI are rarely reported.

Although this case is an isolated instance, it is noteworthy due to its complete follow-up data. The patient initially presented with isolated AOI at disease onset, exhibiting symptoms such as abdominal pain and altered bowel habits. Pathological biopsy results indicated acute cryptitis, and treatment with mesalazine resulted in significant symptom relief and improved endoscopic findings. Subsequent symptom recurrence, followed by reinitiation of treatment, also led to noticeable improvements in symptoms and endoscopic appearance. Consequently, based on symptoms, endoscopic findings, histopathology, and treatment response, the patient was diagnosed with UC, and mesalazine treatment proved effective. This case highlights that UC can occasionally present as isolated AOI without other manifestations of gastrointestinal involvement. Clinical symptom variations aligned with endoscopic changes in AOI, suggesting that AOI can serve as an indicator of UC disease activity. The patient has been followed up for over four years with no evidence of other colonic involvement. Future follow-ups will monitor for potential involvement of other colonic segments.

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CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

All authors: contributed to the design of this manuscript. XL wrote the first draft and all the authors have proofread the manuscript.

ETHICAL APPROVAL

Written informed consent was obtained from the patient.

DATA AVAILABILITY STATEMENT

Data available on request from the authors.

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