

Extensive Oral Aphthous Ulceration Leading to the Diagnosis of Fistulizing Crohn's Disease: A Case Report

Running title: Oral Aphthous Ulceration in Fistulizing Crohn's Disease

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Citation: Daniela B, Marta B, Catarina C, Cristiana S, Marta M. Extensive Oral Aphthous Ulceration Leading to the Diagnosis of Fistulizing Crohn's Disease: A Case Report. *Medi Clin Case Rep J* 2026;4(4):2046-2049. DOI: doi.org/10.51219/MCCRJ/Barros-Daniela/537

Received: 13 October, 2026; **Accepted:** 19 October, 2026; **Published:** 21 October, 2026

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A B S T R A C T

Background: Crohn's disease has a heterogeneous presentation and oral manifestations may precede its diagnosis. Severe recurrent aphthous ulceration accompanied by constitutional symptoms and perianal disease warrants investigation for an underlying systemic disorder.

Case presentation: A 20-year-old man presented with two weeks of severe odynophagia and extensive oral aphthous ulceration that impaired oral intake. He reported recurrent oral ulcers over the preceding year, a 20-kg weight loss over one-month, chronic diarrhoea and a recently treated perianal abscess with fistula formation. Investigations revealed microcytic iron deficiency anaemia, elevated inflammatory markers and markedly elevated fecal calprotectin. Imaging showed extensive bowel inflammation, intramural rectosigmoid abscesses and a left intersphincteric perianal fistula. Flexible sigmoidoscopy demonstrated superficial erosions and ulcers and biopsy showed chronic active colitis. Multidisciplinary assessment established a diagnosis of moderate-to-severe Crohn's disease with fistulizing perianal involvement. Systemic corticosteroids, nutritional support and antibiotic therapy were followed by resolution of the oral lesions, recovery of oral intake and progressive weight gain. Outpatient gastroenterology follow-up was arranged for maintenance treatment.

Conclusion: Severe recurrent oral ulceration may be a prominent presenting feature of Crohn's disease. Associated weight loss, diarrhoea, iron-deficiency anaemia and perianal disease should prompt integrated clinical, laboratory, endoscopic and imaging assessment, even when abdominal pain is absent.

Keywords: Crohn's disease; Extraintestinal manifestations; Inflammatory bowel disease; Oral aphthous ulcers; Perianal fistula

Introduction

Crohn's disease (CD) is a chronic, immune-mediated inflammatory bowel disease (IBD) characterized by transmural inflammation that may affect any segment of the gastrointestinal tract from the mouth to the anus^{1,2}. The disease follows a relapsing-remitting course with considerable heterogeneity in location, behavior, severity and therapeutic response¹.

Although the terminal ileum and colon are most commonly affected, isolated colonic and upper gastrointestinal involvement are well-recognized phenotypes. Despite advances in diagnostic methods and treatment, early diagnosis remains challenging because of the wide spectrum of clinical presentations and the lack of a single definitive diagnostic test^{1,2}.

The global incidence and prevalence of Crohn's disease have increased over recent decades, particularly in industrialized countries. The disease most commonly presents during early adulthood and is associated with substantial long-term morbidity and impaired quality of life^{1,2}.

Clinical manifestations vary according to the location and extent of intestinal inflammation. The classic presentation includes chronic diarrhoea, abdominal pain, weight loss, fatigue and low-grade fever. However, up to 40% of patients develop extraintestinal manifestations, which may involve the musculoskeletal, dermatological, hepatobiliary, ocular and oral systems and may occasionally precede gastrointestinal symptoms, contributing to diagnostic delay³⁻⁵.

Oral manifestations-including recurrent aphthous ulcerations, mucosal edema, linear ulcerations, lip swelling and gingival inflammation-represent one of the most frequent mucocutaneous manifestations of IBD. Although recurrent aphthous ulcers are common and usually self-limiting, severe, persistent or recurrent lesions associated with constitutional symptoms should prompt investigation for underlying systemic disorders, including Crohn's disease. Likewise, perianal fistulas and abscesses are characteristic manifestations associated with a more aggressive disease phenotype and poorer long-term outcomes^{4,5}.

The diagnosis of Crohn's disease relies on the integration of clinical, laboratory, endoscopic, radiological and histopathological findings^{1,2}. Early recognition of atypical presentations is essential because prompt treatment may prevent disease progression and irreversible bowel damage⁶.

Case Presentation

A 20-year-old man presented to the Emergency Department with a two-week history of severe odynophagia and extensive painful oral ulcerations that significantly impaired oral intake. One day before admission, he had attended the Emergency Department for the same complaint and was diagnosed with presumed herpetic stomatitis. He was treated with oral acyclovir and topical corticosteroids without clinical improvement. The patient reported recurrent episodes of oral aphthous ulcerations during the preceding year, although none had been as severe as the current episode. He also described an unintentional weight loss of approximately 20 kg over one month, anorexia, intermittent mucous-bloody diarrhoea and progressive fatigue. His medical history was notable for a recent perianal abscess associated with a draining fistulous tract, for which he had received oral antibiotic therapy. He denied chronic abdominal

pain, respiratory symptoms, urinary complaints or recent unprotected sexual intercourse. There was no known family history of inflammatory bowel disease or autoimmune disorders.

Clinical findings

On admission, the patient was alert and hemodynamically stable. His temperature was 37.5°C, blood pressure was 106/72 mmHg, heart rate was 114 beats/min and oxygen saturation was 99% on room air. Physical examination revealed multiple painful aphthous and vesicular lesions with a whitish exudative base and surrounding erythema involving the soft palate, tonsillar pillars, right lateral border of the tongue and lower lip (**Figure 1**). Oropharyngeal erythema was present without airway compromise. A perianal fistulous opening with continuous purulent drainage and surrounding inflammatory changes was identified. No palpable lymphadenopathy, peripheral edema or abdominal tenderness was observed.



Figure 1: Extensive oral aphthous and ulcerative lesions at presentation. Lesions involve the soft palate, tonsillar pillars, tongue and lower lip, as described in the clinical examination. Yellow arrows identify soft-palate ulcerations.

Diagnostic assessment

Initial laboratory investigations demonstrated microcytic iron-deficiency anemia associated with elevated inflammatory markers. Iron studies confirmed iron deficiency, whereas serum vitamin B12 levels were within the normal range. Fecal calprotectin was markedly elevated, indicating significant intestinal inflammation (**Table 1**). Serological testing for human immunodeficiency virus and hepatitis B virus was negative.

Autoimmune investigations showed an elevated erythrocyte sedimentation rate and rheumatoid factor, while antinuclear antibodies and anti-cyclic citrullinated peptide antibodies were negative. Anti- *Saccharomyces cerevisiae* antibodies (ASCA) were negative. Contrast-enhanced computed tomography of the chest, abdomen and pelvis demonstrated diffuse colonic wall thickening involving the distal transverse, descending, sigmoid colon and rectum, together with terminal ileal wall thickening, mesenteric lymphadenopathy and findings suggestive of inflammatory bowel disease. The patient was admitted for further investigation and multidisciplinary management. Flexible sigmoidoscopy revealed diffuse mucosal inflammation characterized by multiple superficial erosions and ulcers (**Figure 2**) and biopsies were obtained for histopathological examination and cytomegalovirus exclusion. The specimen showed lesions

consistent with chronic active colitis. Pelvic magnetic resonance imaging subsequently demonstrated marked inflammatory thickening of the distal transverse colon, descending colon, sigmoid colon and rectum with extensive submucosal edema. Circumferential intramural abscesses involving the distal sigmoid colon and rectum were identified, together with a left intersphincteric perianal fistula extending approximately 5 cm to the skin surface. No intra- abdominal collections were detected.

Table 1: Laboratory findings at admission.

Parameter	Result	Reference range	Units
Hemoglobin	9.6	13.5–17.5	g/dL
Hematocrit	31.0	41–53	%
Mean corpuscular volume	72	80–96	fL
White blood cell count	11.8	4.0–10.0	$\times 10^9/L$
Neutrophils, absolute	8.6	2.0–7.5	$\times 10^9/L$
Platelets	438	150–450	$\times 10^9/L$
C-reactive protein	86.4	<5	mg/L
Erythrocyte sedimentation rate	70	<15	mm/h
Ferritin	18	30–400	ng/mL
Serum iron	24	65–175	$\mu g/dL$
Total iron-binding capacity	412	250–450	$\mu g/dL$
Transferrin saturation	5.8	20–50	%
Vitamin B12	348	200–900	pg/mL
Folate	6.2	>4.0	ng/mL
Albumin	3.2	3.5–5.0	g/dL
Fecal calprotectin	1820	<50	$\mu g/g$

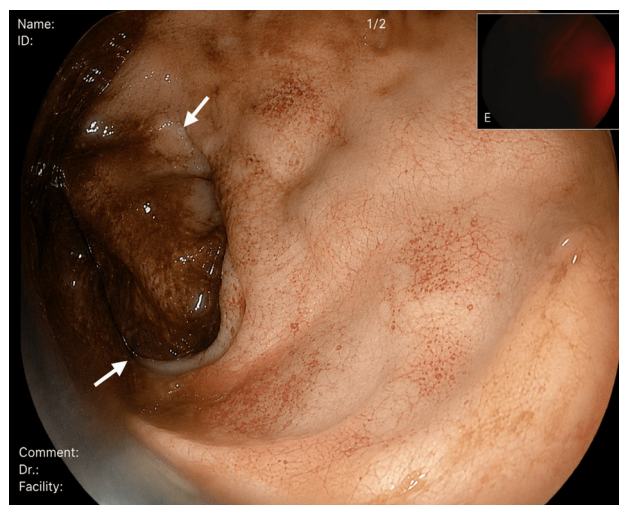


Figure 2: Flexible sigmoidoscopy findings. Diffuse colonic mucosal inflammation with superficial erosions and ulcerations. White arrows indicate mucosal lesions.

Considering the combination of severe colonic inflammation, fistulizing perianal disease, markedly elevated fecal calprotectin, constitutional symptoms and characteristic extraintestinal manifestations, a diagnosis of moderate-to-severe Crohn's disease was established.

Treatment and outcome

The patient initially received empirical intravenous ceftriaxone and metronidazole for suspected perianal infection, together with exclusive parenteral nutritional support because of severe odynophagia and inadequate oral intake. Following multidisciplinary discussion with the Gastroenterology

Department, systemic corticosteroid therapy with oral prednisolone (40mg/day) was initiated. A proton pump inhibitor and calcium/vitamin D supplementation were prescribed for corticosteroid prophylaxis. A structured corticosteroid tapering regimen was planned after clinical stabilization. Clinical improvement became evident within the first days of corticosteroid therapy. The oral ulcerations resolved completely, allowing restoration of adequate oral intake and progressive weight gain. Fever did not recur, inflammatory markers decreased and bowel function gradually improved, with formed stools and only occasional minimal rectal bleeding.

After eight days of intravenous antibiotic therapy, the patient remained clinically and hemodynamically stable and was discharged with tapering oral corticosteroids. He was referred for outpatient Gastroenterology follow-up to initiate maintenance therapy and long-term management of Crohn's disease, including assessment for immunomodulatory or biological treatment after appropriate infectious disease screening.

Discussion

Crohn's disease is a chronic inflammatory bowel disease with a highly heterogeneous clinical presentation. Although chronic diarrhoea, abdominal pain and weight loss represent the classical clinical triad, the disease may initially manifest through extraintestinal features, resulting in delayed recognition and diagnosis. The present case illustrates an uncommon presentation in which extensive oral ulceration and perianal fistulizing disease predominated over gastrointestinal symptoms^{1,2}.

Oral manifestations occur in approximately 20-40% of patients with Crohn's disease and may precede the diagnosis of intestinal disease in a subset of individuals. The spectrum of oral involvement ranges from recurrent aphthous ulceration to mucosal swelling, linear ulcerations, cobble stoning, pyostomatitis vegetans and granulomatous cheilitis^{3,4}. Although recurrent aphthous ulcers are common in the general population, severe, persistent or recurrent lesions associated with constitutional symptoms should prompt investigation for systemic inflammatory disorders. In the present case, the severity of oral ulceration resulted in marked odynophagia, nutritional impairment and substantial weight loss, representing the patient's main reason for seeking medical attention⁴.

An additional remarkable feature was the absence of significant abdominal pain despite extensive inflammatory involvement of the colon demonstrated by cross-sectional imaging and endoscopy. This discrepancy between symptom burden and disease extent highlights the variable phenotype of Crohn's disease and reinforces the importance of maintaining a high index of suspicion in young patients presenting with unexplained weight loss, chronic diarrhea, iron-deficiency anemia and extraintestinal manifestations^{1,2}.

Perianal fistulizing disease is one of the most characteristic manifestations of Crohn's disease and is associated with a more aggressive disease phenotype, increased hospitalization rates and a greater likelihood of requiring biological therapy and surgical intervention. The coexistence of a draining intersphincteric fistula, intramural rectosigmoid abscesses and markedly elevated fecal calprotectin strongly supported the diagnosis of Crohn's disease, even though magnetic resonance imaging reported predominantly colonic disease without clear

small-bowel involvement^{6,7}. Indeed, isolated colonic Crohn's disease is a recognized disease phenotype and should remain within the differential diagnosis when transmural inflammation and perianal disease are present¹.

The diagnostic process required integration of clinical presentation, laboratory findings, imaging studies, endoscopic evaluation and multidisciplinary assessment. No single investigation was diagnostic in isolation. Although serological markers such as ASCA may support the diagnosis in selected patients, their limited sensitivity and specificity preclude their use as standalone diagnostic tools². Conversely, fecal calprotectin proved to be a valuable non-invasive biomarker of active intestinal inflammation and correlated well with disease severity⁸.

The patient demonstrated rapid clinical improvement following initiation of systemic corticosteroid therapy combined with nutritional support and antibiotic treatment for septic perianal complications. Resolution of the oral lesions and restoration of oral intake occurred within days, highlighting that the oral manifestations reflected active systemic inflammation rather than isolated oral disease. Current international guidelines recommend early risk stratification in patients presenting with poor prognostic factors, including young age at diagnosis and fistulizing perianal disease, as these individuals frequently require early introduction of immunomodulators or biological therapy to achieve sustained remission and reduce long-term complications^{6,7}.

This case emphasizes the importance of adopting a multidisciplinary approach when evaluating young patients with severe recurrent oral ulcerations, constitutional symptoms and perianal disease. Recognition of these atypical manifestations may facilitate earlier diagnosis of Crohn's disease, allowing prompt initiation of appropriate therapy before irreversible bowel damage develops.

Conclusion

This case highlights an unusual presentation of Crohn's disease in which severe oral aphthous ulceration, profound weight loss and fistulizing perianal disease preceded the establishment of the diagnosis. Despite the absence of prominent abdominal pain, the combination of constitutional symptoms, chronic diarrhea, iron-deficiency anemia and extraintestinal manifestations prompted further investigation, ultimately revealing extensive inflammatory bowel disease. This report emphasizes the importance of maintaining a high index of suspicion for Crohn's disease in young patients presenting with persistent or recurrent oral ulcerations associated with systemic symptoms. Early multidisciplinary assessment and timely initiation of appropriate therapy may facilitate prompt diagnosis, improve clinical outcomes and reduce the risk of disease progression and long-term complications.

Declarations

Patient Consent obtained.

Ethics approval

Ethics approval or exemption status: obtained.

Conflicts of interest

The authors declare no conflicts of interest, no relevant financial relationships within the preceding three years and no other relationships or activities that could have influenced this work.

Funding

No financial support was received from any organization for this work.

Author contributions

Daniela Barros, Marta Barrigas, Catarina Coelho, Cristiana Sousa and Marta Madarnas contributed to concept and design; acquisition, analysis or interpretation of data; drafting; critical revision for important intellectual content; and supervision. All authors reviewed the final version and agreed to be accountable for all aspects of the work.

Acknowledgments

No additional acknowledgments were supplied.

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