# Inflammatory Bowel Disease Presenting as Recurrent Aphthous Ulcers in a 10-Year-Old Boy

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A 10-year-old boy presented with his parents to our pediatric primary care clinic with multiple long-lasting, recurrent oral ulcers and periumbilical abdominal pain after meals several times a week.

## History

The 4 oral ulcerations were first noticed 3½ weeks prior to the clinic visit. He has had oral ulcerations in the past that have occurred since approximately age 3 years. These were evaluated by a dentist and his prior primary care provider, who had diagnosed aphthous ulcers. However, the more recent ulcerations seemed deeper and lasted longer (multiple weeks) compared with the patient's prior ulcerations. The ulcers had initially caused pain with eating and drinking, but this had improved by the time he visited our clinic.

His abdominal pain had been occurring since age 4 years. It was reported as intermittent and short lasting, for 10 to 15 minutes, and typically occurred after meals. He had not reported any vomiting, diarrhea, constipation, weight loss, or bloody stools. He had a medical history of prematurity and was born as a twin at 25 weeks' gestation. He also has a history of eczema.

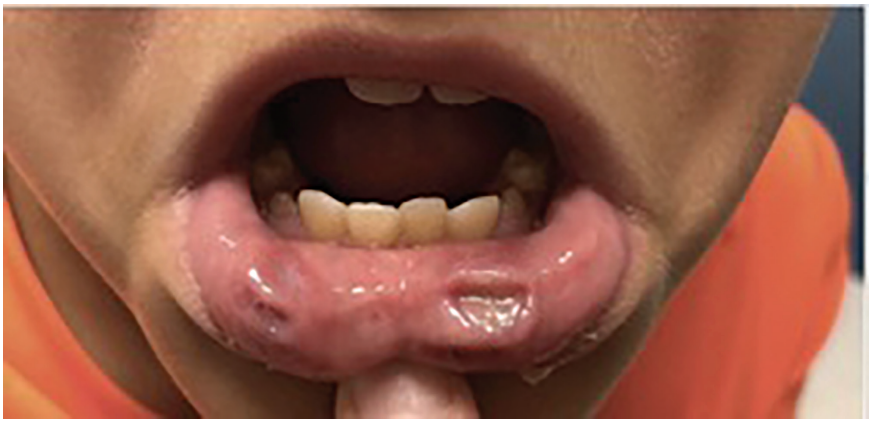

**Figure 1**. Gross image of the oral ulcerations on the inside of the lower lip.

He is not on any medications other than triamcinolone ointment as needed for his eczema. There is no family history of Crohn disease or ulcerative colitis, but his younger sister recently received a diagnosis of celiac disease. His mother has a history of episcleritis, and his cousin has juvenile dermatomyositis.

## Physical examination

He was well-appearing and nontoxic. He had 2 deep and 2 shallow ulcerations on the oral mucosa on the inside of the lower lip (Figure 1). The deep ulcerations had raised borders with a pseudomembrane. The remainder of the oropharyngeal examination was unremarkable.

He had no cervical adenopathy. His lungs were clear to auscultation bilaterally, and he had regular heart rate and rhythm without murmurs upon cardiac examination. He had normoactive bowel sounds, and his abdomen was soft, nondistended, and nontender. No hepatosplenomegaly was noted.

## Diagnostic testing

He was referred to our pediatric gastroenterology team for further testing. Results

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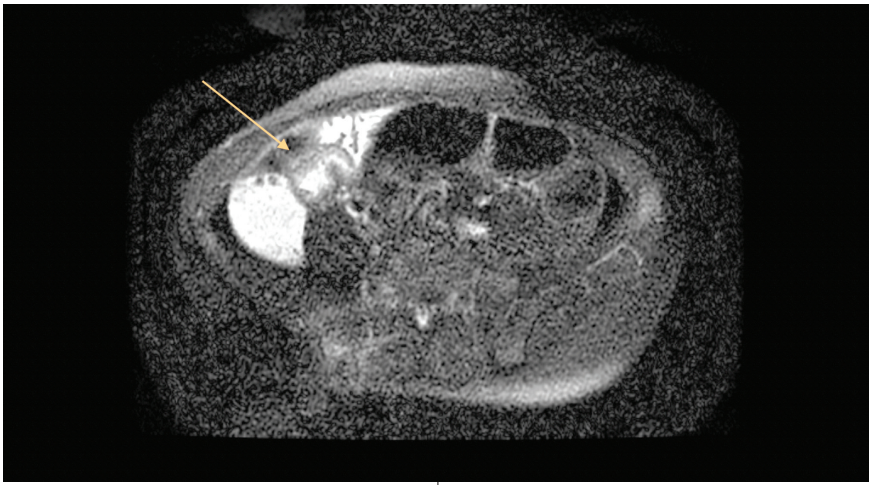

**Figure 2.** A magnetic resonance enterography of the abdomen and pelvis revealed focal enhancement and thickening of a 4-cm segment of terminal ileum extending from the ileocecal valve and edema on the bowel wall.

showed an elevated stool calprotectin level of 153 µg/g (reference range, < 50 µg/g), and after being retested 2 months later, it had increased even more to 372 µg/g.

Celiac disease testing was conducted, results of which showed normal levels of immunoglobulin A (IgA) and immunoglobulin G (IgG), tissue transglutaminase IgA and IgG, and Gliadin IgA and IgG. Results of an endomysial antibodies test were negative.

Test results were within normal limits for complete blood cell count with differential, erythrocyte sedimentation rate, C-reactive protein, thyroid-stimulating hormone, and free T4. Results of a comprehensive metabolic panel were also within normal limits.

An endoscopy and a colonoscopy were then performed, results of which showed normal gross mucosa throughout the esophagus, stomach, duodenum, ileum, and colon. Biopsy results were also negative for diagnostic alterations other than mild chronic inactive gastritis of the gastric antral mucosa and nonspecific reactive changes of the terminal ileal mucosa. A pediatric rheumatologist evaluated the patient and did not find evidence of an underlying rheumatologic disease.

Subsequent magnetic resonance enterography of the abdomen and pelvis revealed focal enhancement and thickening of a 4-cm segment of terminal ileum extending from the ileocecal valve and edema on the bowel wall, thought to be concerning for inflammatory bowel disease (Figure 2).

## Discussion

Aphthous ulcers are common and often are self-limited. However, it is important for providers to be able to differentiate between common aphthous ulcers and the more-severe forms associated with systemic disease.

Recurrent aphthous stomatitis is the most common oral ulcerative pathology, although the etiology is often unknown.[1] Possible inciting factors include trauma, nutritional or microbial causes, stress, and allergies.[2] It is also possible that there is a genetic component to recurrent aphthous stomatitis, as many patients have a family history of the disease with earlier development of disease and increased severity in these cases.[3] These ulcers are often painful, occurring on nonkeratinized mucosa, well-circumscribed, shallow, surrounded by an erythematous border, and covered by a pseudomembrane.[1] This pseudomembrane often consists of fibrin.[4] They can present as single or multiple ulcerations.[4] The ulcerations can vary in size from 1 mm to more than 1 cm.[1] Recurrent aphthous stomatitis is classified into minor, major, or herpetiform types based on size, duration, and scarring potential.[2] Typical treatment for these ulcers is supportive but not curative and includes pain management (either oral or with local anesthetics), antimicrobial mouthwash, and hydration.[1,4] Systemic therapy can include the use of short-term corticosteroids for temporary treatment and anti-inflammatory medications such as dapsone or colchicine for maintenance.[3]

In the pediatric population, the differential diagnosis for chronic or recurrent aphthous ulcers is broad and includes diseases such as recurrent aphthous stomatitis, both viral and bacterial causes such as recurrent herpes infections (often distinguished by the location on hard palate or gingiva), Behçet disease (often associated with genital lesions), periodic fever, aphthous stomatitis, pharyngitis, cervical adenitis syndrome, inflammatory bowel disease, celiac disease, juvenile bullous pemphigoid, childhood linear IgA disease, hereditary epidermolysis bullosa, juvenile dermatitis herpetiformis, cyclic neutropenia, systemic lupus erythematosus, metabolic deficiencies, and HIV/AIDS.[1,4]

Patients with inflammatory bowel disease can present with oral and perioral lesions.[5] These can be present before diagnosis, occur during the illness course, or result from nutritional deficiencies related to the disease.[5] The prevalence of such lesions is variably reported but tends to be more common in individuals with Crohn disease and in the pediatric population.[5] One study found evidence of oral Crohn disease in 41.7% of pediatric patients.[6]

A hypothesized mechanism behind this increase in ulcers in pediatric patients with Crohn disease is that the buccal epithelium is more immunologically active.[5] It is possible for the oral manifestations of Crohn disease to manifest prior to abdominal symptoms.[7] An oral biopsy can be performed to determine whether the characteristic granulomatous inflamma-

tion is present on histopathology.[8] While certain oral lesions are specific to Crohn disease (granulomatous cheilitis) and ulcerative colitis (pyostomatitis vegetans), recurrent aphthous stomatitis is nonspecific.[5] Given the high rates of recurrent aphthous stomatitis in the general population, this can make differentiation difficult. However, the aphthous ulcers present in patients with Crohn disease are often more severe and widespread.[5]

## Patient outcome

Our patient was started on colchicine by the pediatric rheumatologist and has noted some improvement with the ulcers. The pediatric gastroenterologist elected not to begin treatment for inflammatory bowel disease. As for now, the patient's abdominal pain has resolved, he does not have any growth failure, and his colonoscopy did not show evidence of inflammatory bowel disease.

There are few studies that have evaluated how to interpret normal colonoscopy results in the setting of elevated fecal calprotectin levels. One study group looked at the outcomes of 67 patients (although the study did not specify the ages of the patients) with abnormal fecal calprotectin levels followed by normal endoscopic examinations.[9] They found that patients with an initial fecal calprotectin level of more than 225 µg/g and subsequent elevated levels were more likely to have organic disease at follow-up.

However, another study group who evaluated patients with elevated fecal calprotectin and colonoscopy results within normal limits did not find an increased risk for developing gastrointestinal disease.[10] In our patient's case, because of a high suspicion that he may go on to develop clinically significant Crohn disease, the pediatric gastroenterologist is planning to follow-up closely for any changes and is considering further evaluation with capsule endoscopy and repeat fecal calprotectin.

### References


1. Messadi DV, Younai F. Aphthous ulcers. *Dermatol Ther*. 2010;23(3):281-290. https://doi.org/10.1111/j.1529-8019.2010.01324.x
2. Akintoye SO, Greenberg MS. Recurrent aphthous stomatitis. *Dent Clin North Am*. 2014;58(2):281-297. https://doi.org/10.1016/j.cden.2013.12.002
3. Cui RZ, Bruce AJ, Rogers RS 3rd. Recurrent aphthous stomatitis. *Clin Dermatol*. 2016;34(4):475-481. https://doi.org/10.1016/j.clindermatol.2016.02.020
4. Stoopler ET, Al Zamel G. How to manage a pediatric patient with oral ulcers. *J Can Dent Assoc*. 2014;80:e9. https://jcda.ca/article/e9
5. Katsanos KH, Torres J, Roda G, Brygo A, Delaporte E, Colombel JF. Review article: non-malignant oral manifestations in inflammatory bowel diseases. *Aliment Pharmacol Ther*. 2015;42(1):40-60. https://doi.org/10.1111/apt.13217
6. Harty S, Fleming P, Rowland M, et al. A prospective study of the oral manifestations of Crohn's disease. *Clin Gastroenterol Hepatol*. 2005;3(9):886-891. https://doi.org/10.1016/s1542-3565(05)00424-6
7. Coenen C, Börsch G, Müller KM, Fabry H. Oral inflammatory changes as an initial manifestation of Crohn's disease antedating abdominal diagnosis. Report of a case. *Dis Colon Rectum*. 1988;31(7):548-552. https://doi.org/10.1007/bf02553730
8. Padmavathi B, Sharma S, Astekar M, Rajan Y, Sowmya G. Oral Crohn's disease. *J Oral Maxillofac Pathol*. 2014;18(Suppl 1):S139-S142. https://doi.org/10.4103/0973-029x.141369
9. Mohammed N, Smale S. PTU-125 positive calprotectin but negative investigations-what next? *Gut*. 2012;61(Suppl 2):2-A36. https://gut.bmj.com/content/61/Suppl_2/A236.2
10. Hovstadius H, Lundgren D, Karling P. Elevated faecal calprotectin in patients with a normal colonoscopy: does it matter in clinical practice? A retrospective observational study. *Inflamm Intest Dis*. 2021;6(2):101-108. https://doi.org/10.1159/000513473