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# Chronic Peri-Oral Dermatitis as the First Manifestations of Crohn's Disease

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## Introduction

- Crohn's disease (CD) is a chronic inflammatory disease of uncertain etiology, which can involve any part of the gastrointestinal tract from mouth to the perianal area.
- Oral manifestations have been reported in CD. Oral manifestations of CD may coincide with the intestinal manifestations, or less commonly precede them.
- It is not clear whether the presence of these manifestations is a marker of active disease, but their recognition can constitute important clue for diagnosis and management of CD.

## Methods

- Case details and clinical data were obtained from the electronic medical records
- Images were obtained from the electronic medical records and were requested from the pathology department following the routine protocol
- Literature search was done using PubMed and Medline data bases using the following key words: Crohn's disease, oral, dermatitis.

## Case presentation

- An 18-year-old female with no significant history presented to her primary care physician with recurrent lip swelling and dryness for the last 3 years.
- She had thought initially these symptoms due to mild allergy and was diagnosed with angioedema without urticaria.
- Her exposure history was inconsistent and testing for hereditary angioedema, including C1 esterase and C4 levels were unremarkable.
- She was referred to a dermatologist who diagnose her with peri-oral dermatitis.
- Treatment with antihistamines, topical fluocinolone, triamcinolone, and petroleum jelly-based products were all unsuccessful. She was treated with oral prednisone for a presumed pemphigus vulgaris with a symptomatic improvement, but she did relapse two weeks after weaning steroids.
- Due to chronicity of symptoms, a lip biopsy was ordered and showed a psoriasiform dermatitis with a granulomatous inflammation. Direct immunofluorescent testing was unremarkable.
- These results were concerning for CD versus cheilitis granulomatosa. By that time, she denied gastrointestinal symptoms, weight loss or family history of inflammatory bowel disease.
- Further testing revealed a normal ferritin, vitamin B12, folate, antineutrophil cytoplasmic antibodies, and QuantiFERON-TB.
- She was referred to gastroenterology for further evaluation. An esophagogastroduodenoscopy showed a normal esophagus, stomach and examined duodenum. Biopsies from stomach and duodenum were unremarkable. A colonoscopy showed a normal examined perianal area, colon and terminal ileum. Biopsies from the left colon showed a focal active colitis, foci of neutrophilic cryptitis and focal epithelioid granuloma without dysplasia. Stains for acid fast bacilli and fungi were negative. Biopsies from the right colon and terminal ileum were unremarkable.

## Case presentation, con't

- A magnetic resonance enterography showed no evidence of an active small bowel CD.
- Given the early onset and wide spread distribution, the decision was to start adalimumab 40 mg subcutaneously every 2 weeks.
- Her oral disease improved significantly.
- A repeated colonoscopy with biopsies was unremarkable.

## Images



Figure 1 (A, B): Peri-oral dermatitis.

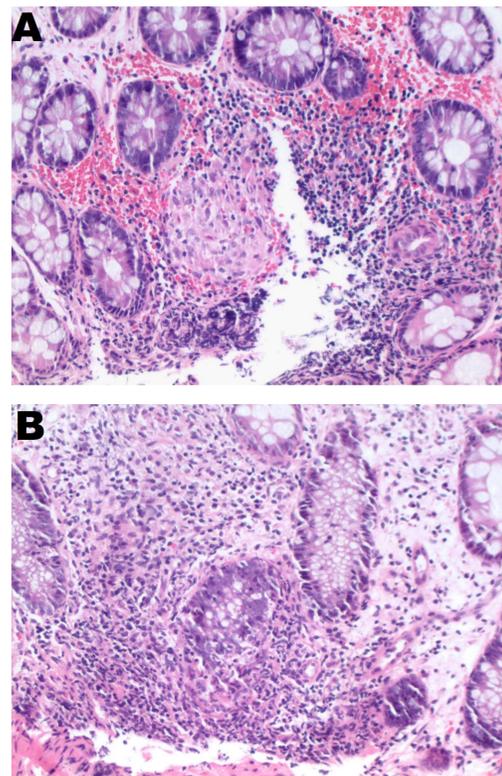


Figure 2: Left colonic biopsy. A) shows small epithelioid granuloma. B) shows the single focus with neutrophilic cryptitis.

## Discussion

- Extraintestinal manifestations are not uncommon in CD. They occur in 25-40% of cases and can be the first presentation of the disease.
- Oral manifestations have been reported in both adults and pediatric patients with CD, even-though, they are more common in children. The reported prevalence of these manifestations varies widely between different studies, and range between 0.5% - 50% in adult and 10% - 80% in pediatric patients.
- Oral manifestations of CD may coincide with the intestinal manifestations, or less commonly precede them.
- They are divided into specific, which are related to the same disease process, and non-specific manifestations.
- Aphthous ulcers, angular cheilitis, swelling of the lips, cheeks and gingiva, cobblestoning of the mucosa, deep linear ulcers, mucosal tags and periodontal disease have been reported in the literature.
- It is not clear whether the presence of these manifestations is a marker of active disease, but their recognition can constitute important clue for diagnosis and management of CD, especially that an isolated oral disease is uncommon as a first presentation of the disease.
- The first step in treating these oral manifestations is to evaluate for, and control, intestinal disease.
- The mainstay of treatment is usually treatment of CD. Both systemic as well as topical treatment has been used.

## Conclusions

- Oral manifestations, even though they are well reported in association with CD, they are uncommon to be the initial presentation of the disease.
- Infections, nutritional deficiencies and medication side effects are important to consider as differential diagnosis.
- Chronic oral disease, especially with an evidence of orofacial granulomatosis prompt a further evaluation for an intestinal disease.
- A careful evaluation of the oropharynx is an integral part of the physical examination of CD patients.

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