

## SHORT COMMUNICATIONS

### Crohn's Disease Presenting as Isolated Lower Lip Swelling

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#### CASE REPORT

A woman in her late 20s presented with a 6-month history of lower lip swelling and painful oral mucosal lesions. History was significant for asthma, bipolar disorder, headaches, and a history of gastrointestinal disease (records were requested to verify a diagnosis). Medications included albuterol, quetiapine, lamotrigine, and ibuprofen. Review of systems was significant for occasional abdominal cramping. There was no family history of similar facial swelling. Clinical examination revealed lower lip edema, cheilitis, and two oral aphthous ulcerations without evidence of facial palsy (Figure 1).

Laboratory evaluation revealed negative antinuclear, anti-double-stranded DNA, anti-smith, anti-ribonucleoprotein, anti-SCL-70, anti-SSA, and anti-SSB antibodies. C1 esterase inhibitor was 34 mg/dL (ref 21-39 mg/dL), functional C1 inhibitor was 100% (normal >68%), C3 was 116 mg/dL (ref 90-180 mg/dL), and C4 was 27 mg/dL (ref 16-47 mg/dL). C-reactive protein (CRP) was 0.70 (ref 0-0.70 mg/dL) and erythrocyte sedimentation rate (ESR) was 16 mm/h (ref 0-20 mm/h). Angiotensin-converting enzyme (ACE) was 62 U/L (ref 9-67 U/L) and vitamin D was 25 ng/mL (ref 30-150 ng/mL). Quantiferon Gold was negative.

A punch biopsy of the inner lower lip demonstrated an inflammatory infiltrate of lymphocytes and plasma cells with non-caseating collections of histiocytes, consistent with granulomas (Figure 2). No refractile material was identified under polarized light. Outside records verified a diagnosis of Crohn's disease as identified on endoscopy/colonoscopy at age 16. At that time, she was treated with corticosteroids for four years with subsequent remission and no further follow up.

**Figure 1.** Initial clinical presentation of Crohn's disease with lower lip protrusion and edema.



We attempted treatment with intralesional corticosteroids, however, the patient tolerated less than 1cc of 5 mg/cc triamcinolone and declined further treatment secondary to pain. She was referred to gastroenterology for endoscopy and colonoscopy; no active Crohn's disease was identified.

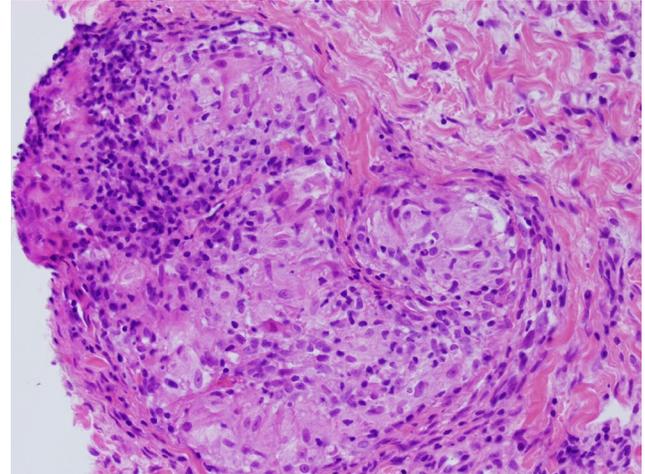
## DISCUSSION

First described by Dudeney *et al.* in 1969, the oral manifestations of Crohn's disease include specific findings: cobblestoning, labial and buccal swelling with vertical fissures, mucosal tags, and granulomatous cheilitis, as well as non-specific findings: aphthous ulceration and angular cheilitis.<sup>1-3</sup> In a study of 228 oral Crohn's lesions, lip involvement was the most common (57), followed by gingiva (40), vestibular sulci (31), and buccal mucosa (25).<sup>3</sup> In rare cases, oral manifestations may be the initial and/or only presentation of disease.<sup>4</sup>

Histologically, Crohn's disease presents with non-necrotizing granulomatous inflammation. Differential diagnosis includes sarcoidosis, foreign body reaction, and infections such as tuberculosis, cat-scratch disease, histoplasmosis, etc.<sup>5</sup> Miescher cheilitis and Melkersson-Rosenthal syndrome are other key differential diagnoses presenting with idiopathic lip edema, induration, angular cheilitis, and granulomatous inflammation. Melkersson-Rosenthal syndrome also presents with facial palsy, unilateral edema, and tongue fissures. Additional pathologies can present with lip edema without granuloma formation, including drug-induced angioedema, C1 esterase inhibitor deficiency, and dietary allergies, including cinnamon and benzoate.<sup>6</sup>

Treatment depends on disease location and severity; thus, endoscopic evaluation is

**Figure 2.** Histopathological analysis of punch biopsy specimen of the left lower lip showing non-necrotizing granulomatous inflammation, consistent with Crohn's disease (Original magnification x 20).



warranted. However, oral manifestations may occur in the absence of lower gastrointestinal disease. For isolated oral Crohn's disease, treatment includes topical corticosteroids or tacrolimus, corticosteroid mouthwashes, or intralesional corticosteroids. Systemic treatment with corticosteroids or azathioprine can be used when topical treatments fail, though in this modern era one might consider one of the newer biologic agents.

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