

BRIEF ARTICLES

Granulomatous Cheilitis: A Case Report

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ABSTRACT

Introduction: Granulomatous cheilitis is a rare condition, with an unknown etiological pathway, resulting in inflammation of the lips. This case report demonstrates the efficacy of intralesional triamcinolone with this persistent disorder.

Case Report: A 19-year old man presented with orofacial swelling on the right side for approximately 18 months. Previous treatments of minocycline and metronidazole were unsuccessful. The patient has no other symptoms and an unremarkable family history. A regiment of topical anesthetic benzocaine gel and intralesional triamcinolone showed significant improvement.

Discussion: This rare condition has many possible contributory factors with very few established treatments. Our patient appeared to present either granulomatous cheilitis or monosymptomatic Melkersson-Rosenthal syndrome. Our primary goal was to abate the patient's symptoms for their comfort, hence the implication of the benzocaine gel.

Conclusion: There are various therapeutic methods described, however we believe intralesional triamcinolone injections with prior application of topical anesthetic to ease discomfort, could be an alternative treatment.

INTRODUCTION

Granulomatous cheilitis is an inflammatory disorder characterized by swelling of the lips.¹⁻⁴ The etiology of granulomatous cheilitis is unclear.¹⁻² The inflammation is generally painless and may be intermittent initially, but tends to persist over time.¹ Due to the inconsistency of the presentation of

symptoms over time and the difficulty in isolating an underlying mechanism a final diagnosis of granulomatous cheilitis may only be rendered after a few years.⁴ Granulomatous cheilitis is considered a rare entity, with only a few cases reported in the literature.² We report a case of biopsy proven granulomatous cheilitis in a young Caucasian man

successfully treated with triamcinolone intralesional injection.

CASE REPORT

A 19-year-old Caucasian man presented with swelling of the upper lip, primarily on the right side. The swelling had been persistent for roughly 18 months. There was no facial and other oral swelling. He had no other systemic symptoms, including respiratory and gastrointestinal. He had no personal or family history of Crohn's disease. He also had no history of atopy and no therapy with an ACE inhibitor or calcium channel blocker. Upon physical examination there was asymmetrical edema of the right side of upper mucosal lip (Figure 1). A skin biopsy from his right side of upper mucosa lip revealed collections of histiocytes in the dermis and slight spongiosis that are consistent with granulomatous cheilitis (Figure 3). PAS and Fite stains failed to reveal microorganisms. He underwent pin-prick testing and North-American panel patch testing which were all negative. Chest X-ray was negative for sarcoid. During his visits, he repeatedly mentioned how distressed he was with his condition. He was treated with minocycline 100 mg BID and metronidazole 500mg BID for one month with no improvement. He had minimal improvement with intralesional injection of 10mg/cc triamcinolone but complained about the pain. We then decided to apply 10% benzocaine gel before injecting him with 20mg/cc triamcinolone. After 5 injections every two weeks, he had significant improvement (Figure 2).

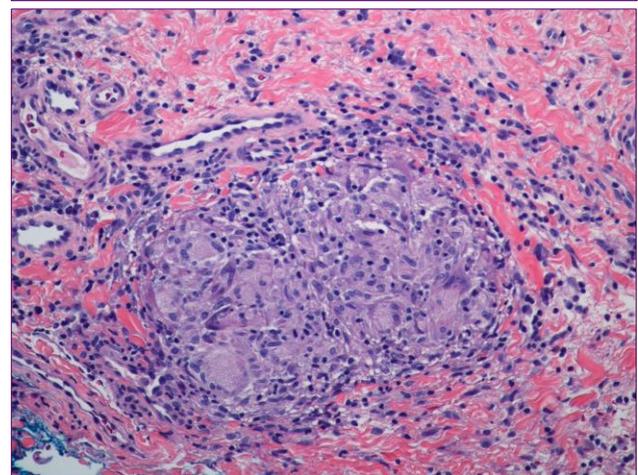
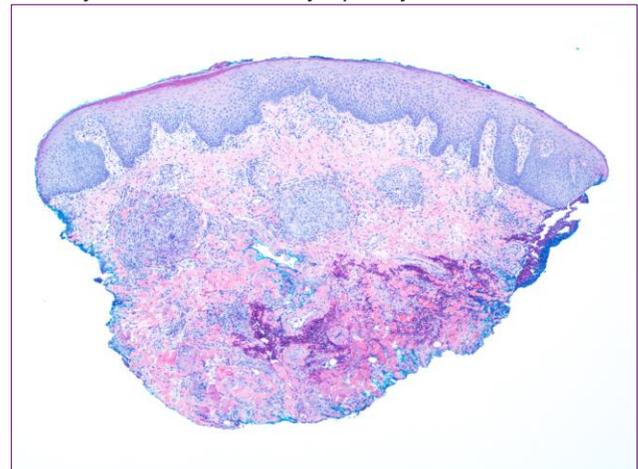
Figure 1. Patient at his initial presentation to our clinic.



Figure 2. Patient after five sessions of treatment.



Figure 3. Nodular aggregates of epithelioid histiocytes with admixed lymphocytes.



DISCUSSION

Granulomatous cheilitis is a recurrent and persistent asymptomatic swelling of the lips.⁵ It is considered as part of orofacial granulomatosis (OFG), which is an entity describing orofacial swelling caused by noncaseating granulomatous inflammation in the absence of systemic disease.^{2,6} The incidence of granulomatous cheilitis is estimated to be 0.08%.⁵⁻⁷ It has no predilection to sex, age, or race.^{6,7} A cell-mediated hypersensitivity reaction, chronic antigenic stimulation, and cytokine production, all leading to granuloma formation, have been suggested as the etiology of granulomatous cheilitis.⁸ The typical manifestation of granulomatous cheilitis is relapsing and non-tender swelling of the lips that slowly becomes permanent.⁹ It may lead to possible facial distortion that cause significant psychological impact on a young patient.^{6,9}

Granulomatous cheilitis may occasionally present as Melkersson-Rosenthal syndrome (MRS), which is a triad of lip swelling, fissured tongue and peripheral facial nerve palsy.^{3,5} We considered our patient as either purely granulomatous cheilitis or monosymptomatic form of MRS because his symptom is solely the upper lip swelling. The complete triad of Melkersson-Rosenthal syndrome has reportedly present in 8% to 25% of patients, and granulomatous cheilitis is the sole manifestation of the disease in 28% of cases.⁵ Another accompanying disorder of granulomatous cheilitis is the extra-intestinal presentation of Crohn's disease.⁴ The differential diagnosis of persistent lip swelling includes other granulomatous diseases such as sarcoidosis, mycobacterial infection, and histoplasmosis.¹⁰

The management of granulomatous cheilitis is difficult in the absence of knowledge of its etiology. The main purpose of treatment is to improve the patient's clinical appearance and comfort. There is no uniform consistent treatment plan or randomized, placebo-controlled trial that is cited in the literature. However, therapies with anti-inflammatory activity such as corticosteroids and immunomodulatory medications show the most promise.¹ Topical, intralesional, and systemic corticosteroid are all described in literature to be effective for granulomatous cheilitis, with intralesional triamcinolone widely described to be the most effective.^{7,11,12} Immunomodulators that targets tumor necrosis factor (TNF)- α such as adalimumab have been reported to successfully cleared granulomatous cheilitis, although potential adverse effects should always be considered.¹⁰

Our patient had failed systemic treatment with minocycline and metronidazole, but he noticed mild improvement with intralesional triamcinolone. Nonetheless, he was reluctant to have more injections due to discomfort. We then decided to apply 10% benzocaine gel 10-20 minutes prior to injection of 20 mg/cc intralesional triamcinolone. The patient was very satisfied and compliant with this regimen. He was treated every two weeks for a total of five administrations, resulting in complete remission.

CONCLUSION

Granulomatous cheilitis is a rare condition with variable clinical presentation. There are various therapeutic methods described, however we believe intralesional triamcinolone injections with prior application of topical anesthetic to ease discomfort, could be an alternative treatment.

Conflict of Interest Disclosures: None

Funding: None

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