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Case Report

Lip Swelling in a Pediatric Patient: A Case Report and Literature Review

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Abstract

Introduction: Lip enlargement is one of the most challenging problems encountered by physicians and dentists due to the various predisposing factors that are involved.

Case Presentation: Here we report a rare case of recurrent enlargement of both upper and lower lips in a 6-year-old child. The parents reported the presence of recurrent white lesions in the patient's oral cavity since birth. The patient's medical history revealed no known systemic condition and use of any medications. In extraoral examination upper and lower lip enlargement was visible. Incisional biopsy was done and microscopic examination demonstrate. severe mixed inflammatory cell infiltration and numerous eosinophils are scattered in the superficial connective tissue within chronic fungal infection.

Discussion: Candidiasis infection was considered as etiology of lip swelling and patient underwent treatment with systemic antifungal agent, which resulted in complete resolution of the condition. Due to high prevalence of chronic Candida infections, this medical condition should be considered in the differential diagnosis of lip enlargement cases.

Keywords: *Lip Swelling; Lip Enlargement; Chronic Candidiasis Infection; Yeast; Allergic Reaction*

Introduction

Lip enlargement can occur in acute and chronic forms, might be recurrent or persistent in the upper or lower lip or both, and might occur as a result of different etiologic factors, including food and drug hypersensitivity, infection, trauma, neoplasms, systemic conditions and some syndromes such as Melkersson-Rosenthal syndrome [1]. *Candida* infection is one of most important infections affecting the oral mucosa. The diagnosis of *Candida* infections in the oral cavity might be a challenge for clinicians because it is manifested in different forms [2,3]. Previous studies have reported rare cases of concomitant *Candida* infections and lip lesions such as exfoliative cheilitis [4] and erosive lip lesions [5]. In the present case, for the first time we report a recurrent lip enlargement concomitant with chronic *Candida* infection.

Care Presentation

A 6-year-old Irano-Afghan boy, complaining of white lesions in the oral cavity and lip enlargement, was referred to the Department of Oral Medicine, Kerman Faculty of Dentistry by a dermatologist in 2014. The child's parents were unable to determine the exact time when the lips began to enlarge; they reported a long history of recurrent enlargement of the lips. In addition, the parents reported the presence of recurrent white lesions in the patient's oral cavity since birth. The patient reported a mild burning sensation in his oral cavity. Furthermore, the parents reported the presence of similar white lesions in one of the patient's siblings.

The patient's medical history revealed no known systemic condition and use of any medications. The patient had history of urticaria and there were no signs of Gastrointestinal tract problems, facial palsy, and other systemic problem. Patient had meat fewer than one meal in a week. The parents reported a history of skin eruptions with no itchiness on the scalp and the dorsum of the neck and dandruffs. They also complained of the patient's short stature and low weight compared to his peers.

In extraoral examination upper and lower lip enlargement was visible. In addition, the nasolabial fold was indistinct (Figure 1). The vermilion border of the lips was dry and exfoliative. Bilateral angular cheilitis was evident. Intraoral examination of the patient revealed spread white non-keratotic plaques on the lateral borders of the tongue and bilateral buccal mucosa.

Figure 1: *Upper and lower lip enlargement was conspicuous. In addition, the nasolabial fold and philtrum were disappeared. The vermilion border of the lips was dry and exfoliative. Bilateral angular cheilitis was evident.*

Candidiasis infection due to systemic condition, allergic reaction and granulomatosis diseases were considered in differential diagnosis.

The laboratory tests including complete blood count (CBC) with differential count, liver function serum calcium, folic acid , vitamin B_{12} levels, iron deficiency and human immunodeficiency viruses (HIV) tests were ordered for the patient.

The results revealed that hemoglobin concentration (13.4 g/dL), Mean corpuscular volume (MCV) (72.9 FL) and mean corpuscular hemoglobin (MCH) (22.9 Pg) were lower than normal and the Serum iron and the ratio of serum iron to total iron binding capability (SI/TIBC) (16.1) were at borderline. In addition, atypical lymphocytes (6%) were reported. The patient's other test results were normal. Incisional biopsy from lower lip was performed. The pathology laboratory test result was as follows.

The histologic sections exhibit areas of oral mucosa that are covered with parakeratotic stratified squamous epithelium with severe spongiosis. In addition, bacterial colonies are visible. Severe mixed inflammatory cell infiltration and numerous eosinophils are scattered in the superficial connective tissue (Figure 2). Periodic acid-Schiff (PAS) stains show the invasion of *Candida* hyphae to the upper layers of the epithelium (Figure 3).

Based on the biopsy report that showed *Candida* hyphae, candidiasis infection was considered as diagnosis of lip enlargement. systemic antifungal therapy with 100 mg of fluconazole (6 mg/kg) per day for 2 weeks was instituted for eliminating candidiasis as main reason of allergic reaction. In addition, triamcinolone NN was administered for the patient's lip commissures. The patient was referred to a hematologist for the evaluation of anemia, who reached a diagnosis of mild microcytic anemia and initiated treatment with iron supplements, folic acid and vitamin B_{12}. The hematologist considered the possibility of thalassemia minor, which was ruled out with the use of further special diagnostic tests. In addition, the patient was referred to a dermatologist for evaluation of skin lesions. Biopsy of the lesions resulted in a diagnosis of seborrheic dermatitis.

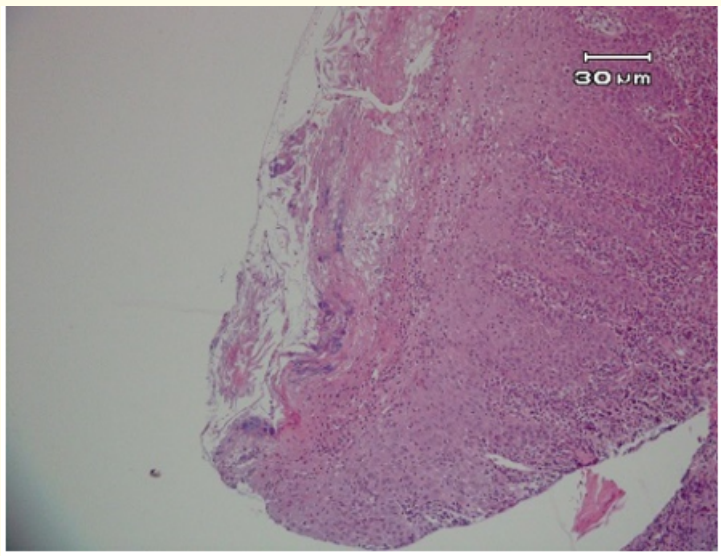

Figure 2: *Lower lip mucosa with severe spongiosis (Hematoxylin-Eosin, original magnification x40).*

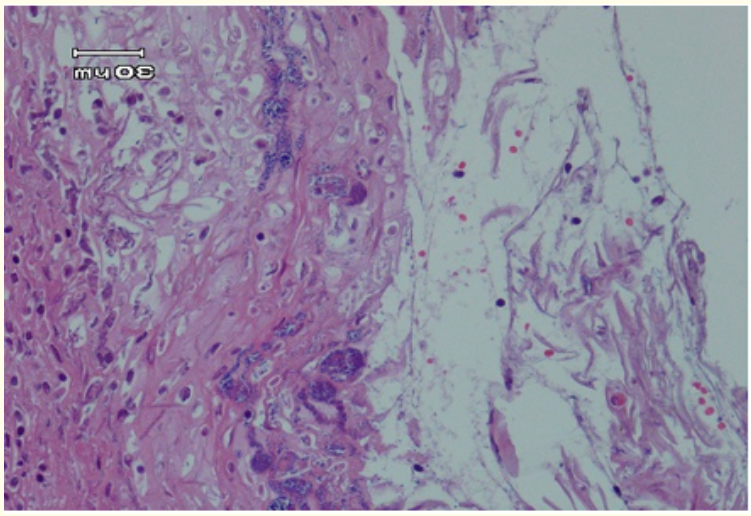

Figure 3: *Invasion of candida hyphae to the superficial layers of the epithelium (PAS staining, original magnification x400).*

The patient was followed on a weekly basis. In the first week, there was no improvement in lip enlargement, intraoral white plaques and the mild burning sensation in the oral cavity. During the second and third weeks after initiation of drug therapy, the patient reported resolution of the burning sensations in the oral cavity; however, there were no changes in lip enlargement and intraoral white plaques. After 4 weeks of fluconazole consumption, lips enlargement, intra oral white plaques and burning sensation were completely resolved (Figure 4). The patient was followed from 2014 to 2017. During the follow ups, there was no recurrence of the oral lesions and lip enlargement. the patient's guardian obtained written informed consent for publication.

Figure 4: *After 4 weeks of consumption of fluconazole lips enlargement, angular cheilitis and lips exfoliative appearance completely resolved.*

Discussion

Lip enlargement might be induced by different etiologic factors, each with its specific treatment modality. Both systemic and local factors can give rise to lip enlargement. Some of the etiologic factors for lip enlargement are food and drug allergies, foreign body reactions, rosacea, vasculitis, infections, cheilitis granulomatosa, Melkersson-Rosenthal syndrome, silica granuloma, oro-facial granulomatosis, Crohn's disease, sarcoidosis, amyloidosis, glandular cheilitis, Langerhans granulomatosis, Wegner's granulomatosis, neoplasms (non-Hodgkin lymphoma, cutaneous T-cell lymphoma, acute myeloblastic leukemia, natural killer/T-cell lymphoma, hairy cell leukemia, and chronic lymphocytic leukemia), chronic idiopathic macrocheilia, Ascher's syndrome. Drug-induced lip enlargement may happen due to the use of cyclosporine, ACE inhibitors and calcium channel blockers. In addition, cases of lip enlargement in systemic lupus erythematosus have also been reported and various references have reported infections as predisposing factors for lip enlargement, including deep fungal infections, mycobacterium, leishmaniasis, leprosy, syphilis, tuberculosis [6-9]. Therefore, it is highly important to thoroughly review the patient's history and be aware of the possible causes that induce this condition in order to be able to diagnose and treat it.

In this case report, the biopsy did not reveal any granulomatous reaction. Also, patient had no history of gastrointestinal sings thus granulomatosis disease such as Crohn's disease and granulomatous cheilitis was ruled out.

Candidiasis is a highly prevalent opportunistic fungal infection of the oral cavity. In children, the condition frequently affects the mucosa. Some factors can predispose to oral candidiasis, including use of drugs (antibiotics, corticosteroids and anti-cancer agents), immune suppression and malnutrition [10].

Daley., *et al.* used the term *Candida* cheilitis in 1995 for the first time to describe the association of *Candida* infection and exfoliative cheilitis [4]. They reported 3 patients with a chief compliant of chronic desquamation of lips without a history of ulceration, swelling, inflammation and hemorrhage, with keratin scales, yeasts and hyphae in microscopic views. The patients underwent local treatment for candidiasis; however, the treatment had no effect on the resolution of the exfoliative process. Daley., *et al.* concluded that keratin scales were an appropriate environment for the growth of *Candida* species and superinfection of *Candida* was probable in exfoliative cheilitis. However, local antifungal treatment was not effective in these patients. Terai., *et al.* in 2006 reported 9 patients with persistent erosion

of the lips, which had not previously resolved with the use of corticosteroids and the presences of *Candida* infection was confirmed after culturing the lesions. All these 9 patients responded to local antifungal treatment and recovered from the condition [5]. These researchers suggested that some cases of cheilitis with unknown etiology that do not respond to local corticosteroid therapy, can be considered as oral manifestation of *Candida* infection.

One of the most important aspects in the successful treatment of *Candida* infection is the identification of the etiologic factors(s). Recurrent and persistent *Candida* infection occurs when the etiologic factor is not successfully eliminated or inadequate treatment is rendered. In this case, all possible causes (diabetes, cyclic neutropenia, immunodeficiency such as HIV, folic acid and B_{12} vitamin deficiency) were ruled out. Patient's Socioeconomic condition raised suspicion of malnutrition and lab tests showed mild iron deficiency. we assumed this can be the main reason for candidiasis infection.

Allergic reactions are one of most common cause of lip enlargement. In this case patient also had diagnosed with seborrheic dermatitis. seborrheic dermatitis is classified as a form of dermatitis, a fungal disease, or an inflammatory disease that can be associated with immune response and inflammation [11]. On the other hand, Bernstein and seidu in 2015 reported 15 case of Chronic vulvovaginal *Candida* hypersensitivity were successfully treated with *Candida* immunotherapy [12]. With histopathologic findings such as severe inflammation and eosinophilia with numerus candidiasis we concluded; hypothetically, lip enlargement can cause by hypersensitivity reaction to candidiasis.

Conclusion

It is for the first time in the literature that a case is being presented about lip swelling caused by *Candida* infection. In the case presented here, one of the challenges for treatment was the identification of the predisposing factor for recurrent *Candida* infection. All the periodic laboratory test results of the patient were normal except for mild anemia. because of limited sources cellular immunology tests could not performed to the diagnosis of immune system dysfunctions which can cause *Candida* infection.

Candida infection is one of the most common infections of the oral cavity, which might be manifested in different clinical forms. In the case reported here, recurrent enlargement was due to chronic *Candida* infection, which completely resolved with antifungal treatment. Since many etiologic factors have been reported for lip enlargement and due to the high prevalence of *Candida* infection in the oral cavity, it is suggested that *Candida* infection be considered in the differential diagnosis of factors inducing lip enlargement.

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