J. Oral Diag. 2020;06:e20210008.

CASE REPORT

Secondary labial cryptococosis in immunocompetent patient: case report

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Abstract:

Cryptococcosis is a fungal infection that produces skin lesions, and more rarely, oral lesions, which are usually secondary to lung and central nervous system involvement. It mainly affects immunosuppressed patients. A 26-year-old patient without comorbidities was diagnosed with central nervous system cryptococcosis, confirmed by detecting fungus in the cerebrospinal fluid, and presenting with an ulcerated single semi-mucosal lip nodule. The anatomopathological study of the lip lesion showed morphology compatible with Cryptococcus spp., confirmed by Grocotts staining. The patient was treated with antifungals and progressed satisfactorily. This is a case report of a rare semi-mucosal lip lesion and its possible origin.

Keywords: Criptococcosis; Oral Manifestations; Lip; Stomatology; Oral Medicine.

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Article received on July 3, 2020.
Article accepted on October 27, 2020.

DOI: 10.5935/2525-5711.20210008

INTRODUCTION

Cryptococcosis is an infection produced by a dimorphic fungus that affects different organs and produces several clinical presentations[1-3]. Common in nature, the main mode of transmission of this pathogen is through the respiratory tract. Autoinoculation is a possible mechanism in single skin lesions[4]. The infection usually develops in immunosuppressed patients, but there are reports of it occurring in immunocompetent patients[5]. The central nervous system (CNS) is commonly primarily affected, and other organs can develop secondary lesions, including surface lesions, following hematogenic dissemination[6]. Skin and oral lesions are, in most cases, secondary[7,8]. There are few reports of primary skin lesions in immunocompetent patients[2,3,5]; thus, the objective of this report is to present a case of semi-mucosal lip lesion associated with CNS cryptococcosis.

CASE REPORT

A 26-year-old male patient with no history of comorbidities presented at the unit complaining of total cranial headache, decreased sensitivity in the left half of the body and a semi-mucosal lesion on the right side of the lip. Computed tomography of the skull showed a nodular formation in the right basal nuclei with adjacent mass effect. The cerebrospinal fluid investigation presented a positive china ink test, with 2% budding and torula [1]. Chest tomography revealed an area of pulmonary atelectasis. Serum tests for other infectious diseases were all negative.The lip assessment was requested after patient's hospitalization. According to the patient, the lip lesion appeared one week after the neurological complaints, and did not present a specific complaint. Examination of the lip lesion showed a sessile nodule with an ulcer-crusted surface, measuring 1 cm in diameter, soft on palpation, and surrounded by an erythematous area (Figures 1 and 2). Ultrasonography of the region showed a lesion with a liquid content of 1.2 cm^3, which may have represented an infectious collection. A complete dermatological examination identified no other lesions. After the initial diagnosis of CNS cryptococcosis was confirmed, the patient underwent a biopsy of the labial lesion; histopathological examination revealed the presence of intracellular yeasts at different stages of development, morphologically compatible with *Cryptococcus* spp. (Figures 3 and 4). Grocott's (Figure 5) and mucicarmine staining confirmed the fungal origin of the lesion. The patient was treated with amphotericin B, flucytosine, and fluconazole, with complete remission of the symptoms and lesions.

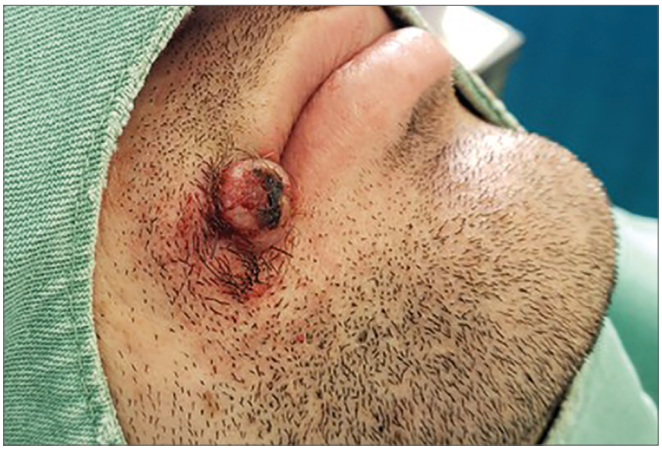

Figure 1. Nodule with ulcer-crusted surface surrounded by erythematous area.

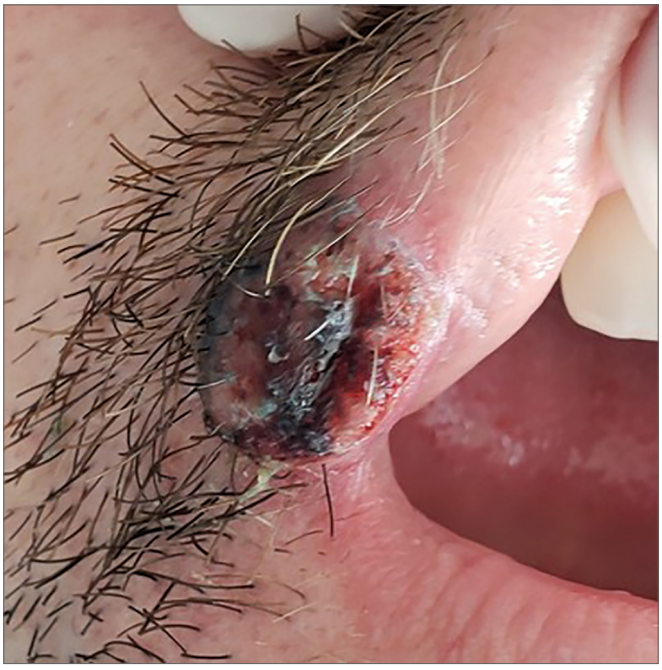

Figure 2. Nodule in more detail. It is a semi-mucosal lip lesion.

DISCUSSION

The clinical presentations of cryptococcosis are related to the immune reactivity of the fungal capsule and immune system capacity [6]. Patients with deficient cellular immunity are more vulnerable to developing an infection and of greater severity, such as in cases of solid organ transplants, autoimmune disease, human immunodeficiency virus-positivity, and chronic corticosteroid users[6,7]. According to some studies, the development of the disease in immunocompetent patients is related to the serotype and genotype of the fungus[2].

Skin lesions are usually secondary to early disease presentations and sometimes considered a marker of

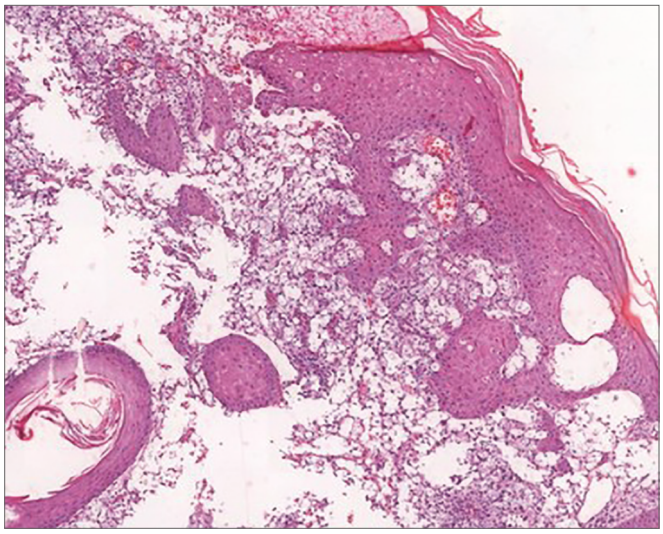

Figure 3. Fragment of squamous mucosa with numerous fungal structures associated with mucinous material and diffuse chronic inflammatory infiltrate (Hematoxylin/eosin, ×20).

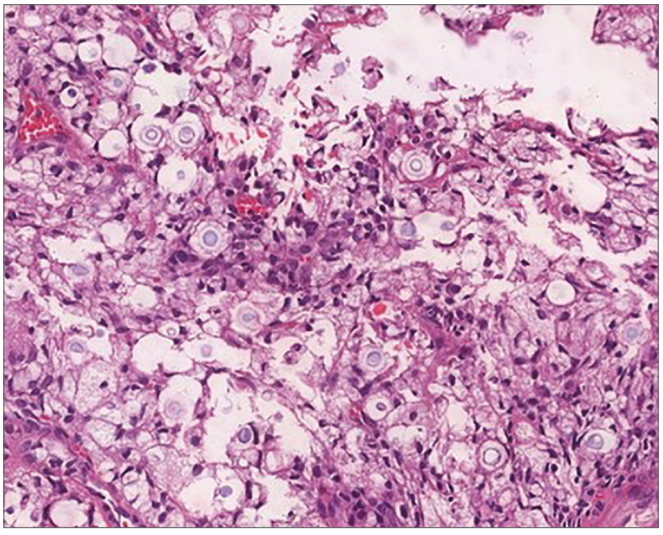

Figure 4. Magnified view of yeasts at different ripening degrees surrounded by mucinous material. Presence of giant cells is also noted. (Hematoxylin/eosin, ×40).

widespread disease[3,5]. They are present in 5–20% of patients[6,7] as multiple lesions, with papular, nodular, acneiform, herpetiform, and pustular clinical aspects. The lesions often resemble molluscum contagiosum[6].

On the other hand, primary cryptococcosis lesions are rarer and have been described in both immunosuppressed and immunocompetent patients. Clinically, these lesions present variably (e.g., acneiform, ulcerated, and necrotic) and may be associated with cellulite and internal gelatinous content[3]. In the present case, there was a single lip lesion with an ulcerated aspect, perilesional cellulite, and liquid content inside. At first, this lip lesion

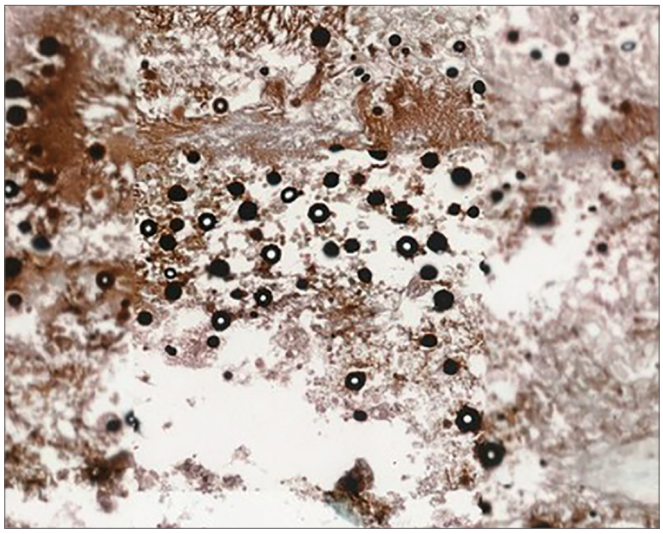

Figure 5. Silver methenamine staining showing intense impregnation in the yeast cell wall (Grocott's, ×40).

could suggest a primary cryptococcosis lesion, possible due to self-inoculation resulting from a local penetrating trauma, as described by Marques et al.[3] A review of the patient's anamnesis identified no history of trauma. According to the criteria set out by Ng & Loo[9], a patient cannot present with injury in another organ to characterize primary cryptococcosis. As the patient presented with CNS cryptococcosis concomitant with the lip lesion, the case could not be classified as one of primary injury. On the other hand, Neuville et al.[1] describe primary cryptococcosis lesions as single ulcerated lesions associated with cellulitis, exactly as observed in the patient in this case report.

Regarding the patient's immunocompetent status, the lip lesion could not be classified as a possible primary lesion. Christianson et al.[4] reviewed 73 cases of primary skin lesion and reported that approximately 50% were in immunocompetent patients. Of these, only 16.4% were in the head and face region.

In light of the above, this case can be considered an isolated secondary presentation of CNS cryptococcosis; however, the fact of being a single lesion in an area susceptible to trauma does not exclude the possibility of self-inoculation. No previous reports of a single semi-mucosal lip lesion in an immunocompetent patient were found in the literature.

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