CASE REPORT

Unique Cytomegalovirus Tongue Ulcer in an Older Adult Patient with Prolonged Systemic Steroid Treatment

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Abstract

Introduction: Cytomegalovirus (CMV) is a member of the Herpesviridae family that infects humans. It infects approximately 90% of adults. It is an opportunistic pathogen, common among immunosuppressed patients and can affect multiple organs. To date, there has been only one reported case of a CMV-related tongue ulcer that occurred during steroid treatment.

Case Presentation: We report a case of CMV tongue ulcer in an elder, immunodeficient patient under prolonged steroid treatment.

Management and Prognosis: A 67-year-old man under chronic steroidal treatment due to severe chronic obstructive pulmonary disease (COPD), with chronic ischemic heart disease (CIHD) and poorly treated diabetes mellitus (DM) was hospitalized in Soroka university medical center. He suffered from an ulcerative wound about 10 mm in its greater diameter with raised margins on the left lateral tongue. Daktarin Oral Gel treatment showed no improvement. On histopathological examination, a diagnosis of CMV-related tongue ulcer was given.

Conclusion: To the best of our knowledge, this is the most detailed case report of CMV tongue ulcer due to prolonged steroid treatment. An early diagnosis of CMV infection in patients with oral lesions is crucial because CMV infection can increase immunosuppression, and is associated with opportunistic infections.

Keywords: Cytomegalovirus, Oral ulcer, Steroid treatment, Tongue, Immunodeficient patient

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Introduction

Cytomegalovirus (CMV) is a member of the Herpesviridae family, in the subfamily Betaherpesvirinae. Human betaherpesvirus 5 known also as human cytomegalovirus or human herpesvirus 5 (HHV-5) is the one that infects humans.[1] CMV is estimated to infect approximately 90% of adults at some point in their lives, this infection is usually subclinical and can persist in this manner for their life.[2] It is an opportunistic pathogen that can affect multiple organs.[3] Opportunistic infections are common among immunosuppressed patients. Cases of CMV oral ulcerations are rare, oral lesions caused by CMV have been reported mostly in transplantation recipients. Human immunodeficiency virus (HIV) patients are also known to suffer from oral ulcers caused by CMV.[4,5] To date, there has been only one reported case of a CMV-related oral ulcer that occurred during steroid treatment.[6] We report a case of CMV tongue ulcer in an elder, immunodeficient patient under prolonged steroid treatment.

Case Presentation

A 67-year-old man was hospitalized in the internal medicine ward due to deterioration in his general condition. Patients' medical records revealed that he was hospitalized several times before the present event. His records revealed a history of respiratory failure due to severe chronic obstructive pulmonary disease (COPD) for which the patient received chronic steroidal treatment with 40 mg Prednisone daily, chronic ischemic heart disease (CIHD) and poorly treated diabetes mellitus (DM). During the previous hospitalization, an Aphthae-like wound on the left side of the tongue about 5 mm in size was noticed. The wound was treated with Daktarin Oral Gel without any improvement. During the present hospitalization, the patient complained about a painful tongue ulcer that was present for about a month and was still growing. On intra oral examination, an ulcerative wound with raised margins was noticed on the left side of the lateral tongue about 10 mm in its greater diameter (Figure

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1). Hard lumps were palpated along the edges of the wound and in its depth, no bite marks and no other signs of chronic trauma were found. An incisional biopsy was performed for a definitive diagnosis and the specimen was sent for a histopathological examination. After the biopsy and more before receiving a result or giving treatment, the lesion showed signs of spontaneous healing. Histopathological findings revealed a deep ulceration with chronic active inflammation, granulation tissue formation and epithelial hyperplasia at the periphery of the ulcer. Massive infiltration by adipose tissue was noticed. Immunostaining for CMV (Dako CCH2+DDG9) was positive in a few cells. Immunostaining for Herpes virus (Bio SB 10A3/BSB-116) was negative. No tumor was seen in this material and no fungi were identified (Figure 3 and 4). Based on these findings, a diagnosis of CMV-related tongue ulcer was given. In a blood test, levels of IgG 133,000 U/ml and IgM 14.4 U/ml, indicated that the patient was previously exposed to CMV, but not recently. The patient was also tested negative for HIV. On follow up, the lesion healed without any anti-CMV medication (Figure 4). The patient passed away shortly after the last follow up.

Discussion

The age-adjusted seroprevalence of cytomegalovirus (CMV) infection is about 50%, however, CMV only rarely presents in oral cavity lesions.[7] A few reports of intraoral CMV infection in the form of nonspecific ulcers on the lips, palate, tongue, and gingiva have been documented.[7,8] Oral lesions caused by CMV have been reported mostly in transplantation recipients and HIV patients, patients with malignant lymphoma, and patients with autoimmune diseases.[2] Ueda in 2013 reported to date the only report of a CMV-related tongue ulcer related to chronic steroidal treatment.[6]

Oral lesions secondary to CMV infection are characterized by deep and painful nonspecific ulceration that is typically solitary. These findings can clinically mimic aphthous or traumatic ulcers. If appearing on the tongue, the lesion may mimic a squamous cell carcinoma (SCC). As soon as these types of ulcers are seen in a patient with prolonged systemic steroid treatment, the involvement of CMV should be considered. Clinical diagnosis of oral ulcers in immunosuppressed patients is often difficult because of various etiologic factors.[4] Biopsy is recommended because the virus and cellular changes are typically only seen deep in the endothelium of the connective tissue and because of the SCC suspicion. A diagnosis of CMV infection can also be confirmed by virus isolation; the presence of antigenemia, which detects polymorphonuclear leukocytes that are positive for the viral antigen pp65.[9]

In the specimen obtained from our patient, isolated CMV

Fig. 1: An ulcerative wound with raised margins on the left side of the lateral tongue. About 10 mm in its greater diameter

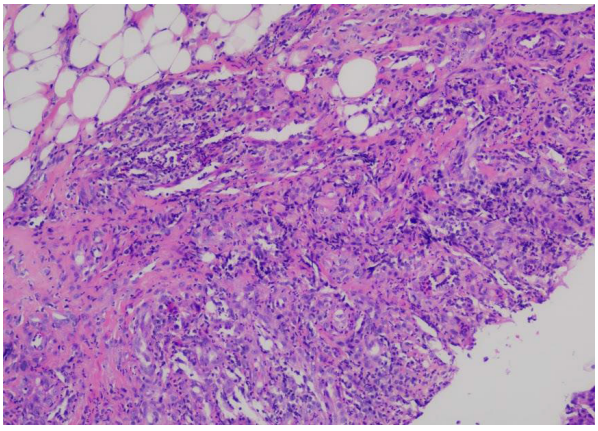

Fig. 2: Ulcerated surface and infiltrating chronic -active interfascicular inflammation. No evidence of cytopathic effect. H& E x10

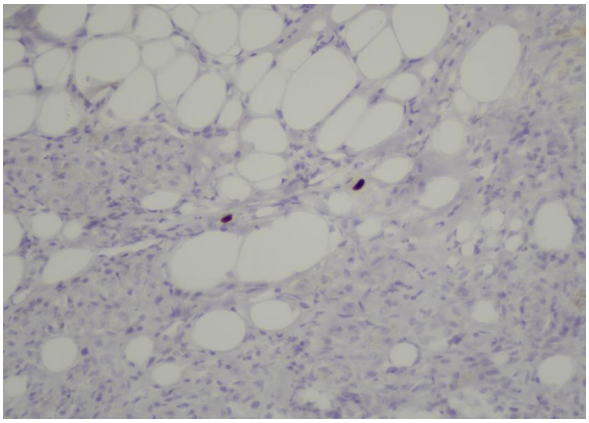

Fig. 3: The same ulcerated region with isolated CMV positive cells CMV stain x20.

Fig. 4: Normal tongue tissue on the follow up

positive cells were detected by immunohistochemical stain within florid nonspecific inflammation; diagnostic cytopathic effect was not evident.

Conclusion

An early diagnosis of CMV infection in patients with oral lesions is crucial because CMV infection can increase immunosuppression, and is associated with opportunistic infections. In conclusion, we report a case of tongue ulcer caused by CMV infection in an older adult patient owing to prolonged systemic steroid treatment for COPD. To the best of our knowledge, this is the second and the most detailed case report of CMV tongue ulcer due to prolonged steroid treatment.

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