A Rare Case of Oral Erythema Multiforme: A Case Report with a Literature Review

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A Rare Case of Oral Erythema Multiforme: A Case Report with a Literature Review

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ABSTRACT

Erythema multiforme (EM) is an acute mucocutaneous hypersensitivity reaction characterized by skin eruptions with or without oral or other mucous membrane lesions. The main two variants are erythema minor and erythema major. Oral disease with typical EM lesions has been suggested as a third variant of EM. Known as oral EM, it is reported less and has no target lesions unlike the other two types, in its primary presentation. **Objective:** To report a manifestation of a rare case of oral EM and discuss various forms of EM including its management. **Case report:** A 22-year-old male patient reported with a complaint of oral and lip ulcers and severe pain for the past 7 days. The patient reported spontaneous onset of the lesions in the form of vesicles after consuming unknown artificially colored food items. The vesicles ruptured within two days leaving ulcers on the lips and the intraoral mucosa, with blood encrustations. The patient was unable to take food, was admitted for hydration, and was kept on corticosteroids. It took around three weeks for the patient to completely recover. **Conclusion:** The positive history of artificially colored food intake followed by the sudden onset of lesions and eruptions on the lips and oral mucosa led us to the diagnosis of oral EM. Early recognition and timely intervention benefits patients because the lesions associated with EM and related disorders can compromise life.

**Keywords:** erythema multiforme, encrustations, vesicles

INTRODUCTION

Erythema multiforme (EM) was first reported by Ferdinand von Hebra in 1866 as an acute and self-limiting disease of the skin and mucous membrane with symmetric scattering of lesions on the extremities and having a typical recurring concentric pattern in the form of target lesions. EM can occur due to certain drug intake or different infections, particularly herpes simplex virus (HSV) infection. Research has implicated certain food preservatives like benzoic acid as etiological factors in certain cases of EM. EM manifests usually in the age group of 20-40 years affecting teenagers and young adults, but it can precipitate as late as 50 years of age or even later. The disease has a male predilection over females in the ratio of 3:2. EM has been classified as EM minor, EM major, Stevens-Johnson syndrome (SJS), and toxic epidermal necrolysis (TEN); EM minor is the mildest form of the disease and TEN is the most severe. EM rarely affects the oral cavity alone thus making it a rare entity. EM with oral mucosa lesions and lip lesions without any skin lesions has been reported by a number of researchers, and this variant has been called oral EM by various investigators. The current study presents a case of oral EM with oral and lip lesions without any skin manifestations and seemingly resulting from a food allergy, which is less reported and hence a rare case.
CASE REPORT

A 22-year-old male patient reported to the department of oral medicine and radiology with a complaint of painful lip ulceration and severe pain for the past seven days. The patient gave a history of attending a party one day prior to the appearance of the ulcers and being served a range of artificially colored food items. There was no significant medical or family history and no recent history of any drug intake. The patient had visited a physician immediately after the appearance of the ulcers and had been prescribed antibiotics and local application of gentian violet. The application of gentian violet had allegedly worsened the condition. Extensive ulceration of the upper and lower lips was noticed on extraoral examination as well as cracks, fissures, and blood encrustations on the lips (Figure 1). Intraoral examination revealed widespread ulceration with yellow bases and erythematous borders on the buccal mucosa, palate, and ventral surfaces of the tongue (Figure 2a, b). There was marked tenderness on palpation. The dorsum of the tongue showed a white coating. The mouth opening was reduced because of marked tenderness around the ulceration and there was increased salivation. There was no previous history of similar ulcers. Based on the history and clinical findings, a provisional diagnosis of EM secondary to unknown food additives was made. The patient was admitted in the constituent hospital for supportive care and hydration as the patient had difficulty eating solid food because of the widespread ulceration. The patient was kept on intravenous saline/ringer lactate at a rate of 100 ml/hour and an oral liquid diet with prednisolone tablets: 10 mg twice daily for the first week; 5 mg twice daily for the second week; and 5 mg once daily for the third week. Tablet paracetamol (500 mg) thrice daily for three days and oral lidocaine gel to facilitate oral intake of food were advised. An antiseptic mouthwash was also prescribed to prevent secondary infection. The patient was evaluated after three days and showed marked improvement of lesions and symptoms. The patient’s hydration level was satisfactory he was discharged with instructions. The patient was also instructed to continue with tablet prednisolone as advised before and to report back after one week. The patient was examined after one week and showed marked improvement in the ulcers on the lips and oral mucosa (Figure 3). Complete remission occurred after three weeks (Figure 4a, b).

DISCUSSION

EM is the result of antigenic challenge evoking allergic host response and manifests as a reactive mucocutaneous disorder. The oral lesions manifest as ves-
icles and bullae that rupture in an explosive manner resulting in widespread sloughing and ulceration of the whole mucous membrane.\textsuperscript{6,11} EM is triggered by various agents, common among them being infections, drugs, and food additives.\textsuperscript{6,11,12} A prior HSV infection has been found to be the precipitating factor in 71% of EM cases.\textsuperscript{13} The current study’s case was the result of food additives based on the history of artificially colored food intake by the patient prior to the onset of the lesions without other relevant history. A large proportion of patients (>50%) have no known cause, and the second largest trigger category is being in an emotional state or stressful conditions.\textsuperscript{10} Although the exact mechanism is not known, it is postulated that the causative agents induce a T-cell mediated immune reaction which results in cytotoxic immunological attack on keratinocytes, which express non-self-antigens, with consequent vesiculation and extensive erosion and blistering.\textsuperscript{13} Anti-epidermal autoantibodies and autoantibodies against desmoplakin I & II have been shown to play a role in a subgroup of EM. EM and similar diseases may involve the humoral immune system in addition to the cellular immune response.\textsuperscript{6,14}

EM can present in several ways ranging from a milder form, EM minor, which is self-limiting, to EM major, Stevens-Johnson syndrome, and TEN, which are progressive and aggressive forms of the disease.\textsuperscript{6,9-12,14} Inflammation of the oral cavity with lesions resembling typical EM lesions was first described by Kenneth in 1968. Such cases, presenting with typical oral EM lesions without any target skin lesions have been categorized as a third variant of EM by several investigators.\textsuperscript{15,16} This variant is considered oral EM and is considered as a chronic condition with recurrences ranging from every three weeks to a single episode yearly. The duration of episodic cycles of oral EM range from 10 days to 42 days.\textsuperscript{8} In the current study’s case, the lesions were restricted to the lips and oral mucosa and did not recur during the first six months of follow-up.

The management of EM depends on the severity. Local lesion care, pain control in the form of topical analgesics or anesthetics, and a softer diet are recommended for milder forms of EM. The milder form generally heals within two to six weeks. For extensive cases, intravenous fluid therapy is recommended to maintain fluid balance.\textsuperscript{17} As the nutritional and hydration status of the current study’s case was compromised, because of the widespread oral and lip lesions, intravenous fluids for two days along with systemic corticosteroid and local application of an anesthetic agent were resorted to.

**CONCLUSION**

Oral EM is considered a rare and less reported type of EM. Oral EM is usually triggered by HSV infections and rarely by adverse drug reaction and food additives. EM of any form, and in particular the oral form, is an incapacitating disease and timely diagnosis, swift management, and proper follow-up is of utmost importance in order to safeguard the recovery phase of patients.

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