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CASE REPORT

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Erythema multiforme complicated with lichenoid mucositis: A case report

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Abstract

Erythema multiforme (EM) is an acute, self-limiting, mucocutaneous disease. While the histopathologic and immunopathologic feature of EM is not highly specific, the diagnosis is mainly based on the etiology and clinical manifestations. This report presented a patient whose clinical investigation indicated EM while histopathology suggested lichenoid mucositis. Considering the classic oral and skin presentation matching EM, as well as the acute onsite and spontaneous resolution of the lesions, the diagnosis highly favored EM. The immunofluorescent findings might suggest a potential common pathologic process on EM and lichenoid mucositis, which requires further investigation.

Keywords: *erythema multiforme, lichenoid mucositis, pathology, diagnosis.*

Introduction

Erythema multiforme (EM) is a mucocutaneous hypersensitivity reaction that can be triggered by infectious agents or medications. Clinically, EM is classified into EM major which presents with extensive involvement of the skin, the oral mucosa and other mucous membranes, which EM minor has less than 10% of skin involvement and minimal to no mucous membrane manifestation. The EM major was ever regarded as a variation of Stevens-Johnson syndrome (SJS) or toxic epidermal necrolysis (TEN); however, this was not fully supported by etiological and pathogenic evidences [1].

The common pathological findings of EM are subepithelial or intraepithelial vesiculation in association with necrotic basal keratinocytes, and mixed inflammatory infiltrate consisting of lymphocytes, neutrophils, and often eosinophils. While the histopathologic and immunopathologic feature of EM is not highly specific, the diagnosis is mainly based on the clinical presentation and the exclusion of other conditions. This report presented a patient whose clinical investigation indicated EM while histopathology suggested lichenoid mucositis.

Case report

A 34-year-old man was referred to our oral medicine clinic for assessment of recurrent oral and skin lesions, occurring 4-5 times per year for over two years. The patient reported an acute onset of multiple oral ulcerations accompanied by hemorrhagic crusting and swelling of the lip that resolved spontaneously in 2-4 weeks. The lesions were confined to oral mucosa in the first several episodes, and then extended to skin and genital mucosa. The patient denied any medical and drug history. During this period, the patient visited the emergency department, ENT clinic and dermatological clinic and was evaluated by the different specialties. Results of the blood tests and ulcer swabs were negative for herpes simplex virus (HSV), human immunodeficiency virus (HIV), venereal disease research laboratory (VDRL) test, and all autoimmune markers. Oral pathergy test was negative. The patient was then diagnosed as Behcet's disease in these clinics and was prescribed with colchicines but with minimal improvement.

Upon our clinical examination, there were diffuse, erythematous, ulcerative lesions on the tongue (Figure 1) and hemorrhagic crusts of the lips (Figure 2). Target lesions were found on the skin of the limbs (Figure 3). The clinical findings were consistent with EM minor. Incisional biopsy was performed on the lower lip and subjected for histopathology and immunofluorescence examination. Histopathologic findings showed a thickened and parakeratotic stratified squamous epithelium with mild patchy non-specific chronic inflammation. The frozen section showed features suggestive of a mild focal lichenoid mucositis. Immunofluorescence examination showed no deposits of IgG, IgM, IgA, C3 or C1q. There is however a strong band of fibrin on the basement membrane at the epithelial mesenchymal junction. The appearances and IF findings suggested lichen planus.

Figure 1. *Diffuse, erythematous, ulcerative lesions on the tongue.*

Figure 2. *Extensive hemorrhagic crusts and ulcerations on the lips.*

Discussion and conclusion

The diagnosis of EM is mainly based on the etiology and clinical manifestations. EM frequently associates with herpes simplex virus, indicating an immunologic event initiated by the virus. Drug is another common triggering agent, which includes nonsteroidal anti-inflammatory drugs, anticonvulsants, sulfonamides, and some antibiotics. The correlation with other immunological conditions like graft-versus-host disease was also proposed but was found to be uncommon [2]. The oral findings of EM range from mild erythema and erosion to large, painful ulcerations and difficulty in drinking and eating. Extensive lip involvement with ulceration and crusting is common. The classic "target" or "iris" skin lesion consists of a necrosis or blister with concentric erythematous rings around it. The diagnosis of EM could be challenging when the causes are identifiable, and it could be more difficult to distinguish from other mucocutaneous diseases with similar presentation when the manifestation is not classical. The acute onset, fast resolution, and self-limiting behavior of EM are the additional challenges in investigation and diagnosis. Clinicians therefore mostly rely on clinical assessment after ruling out more severe conditions with similar manifestations like SJS, TEN, pemphigoid or pemphigus.

Neither medical/drug history nor clinical biochemical investigations suggest the involvement of infection or drug in this patient. The patient could not recall any history of HSV infection, and serology and ulcer swab tests were negative. Although the patients admitted the occasional use of paracetamol and ibuprofen, he could not recall any correlation between the use of the drug and the onset of the lesions. In autoimmune conditions such as pemphigoid or pemphigus, deposition of specific markers like IgG, IgM, IgA, C3 or C1q serve as gold standard of diagnosis, yet they were negative in this case. Interestingly, the immunofluorescent findings of this patient suggest lichen planus, but no lichenoid lesion was found on either the oral mucosa or the skin.

Figure 3. *Target lesions on the skin of the front forearm.*

Considering the classic oral and skin lesions matching the presentation of EM, as well as the acute onsite and spontaneous resolution of the lesions, the diagnosis highly favored EM. The immunofluorescent findings might suggest a potential common pathogenesis and pathologic process on EM and lichenoid mucositis, which might require further investigation [3,4].

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