

Case Report

Herpes associated Erythema Multiforme in Young Child- A Case Report

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ABSTRACT:

Erythema multiforme (EM) is a mucocutaneous disorder that presents clinically as vesicubullous and ulcerative lesions affecting any part of oral cavity when occurring alone in oral cavity. It can commonly be seen on skin too with typical target like lesions. We present a case of EM in a 9 year old girl who complained of recurring lesions on lip. Based on history, clinical presentation and serological investigations, it was diagnosed as herpes associated Erythema multiforme (HAEM).

This case was very well managed with systemic steroids and it highlights the advantage of gentian violet for mucocutaenous lesions and also immunomodulator as maintenance therapy.

KEYWORDS: Erythema multiforme, HAEM, recurrent herpes infection, gentian violet, steroids

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INTRODUCTION:

Erythema multiforme (EM) is an acute, self-limited, inflammatory mucocutaneous disease that manifests on skin and often oral mucosa, although other mucosal surfaces such as the genitalia, may also be involved. EM is considered to be a hypersensitivity reaction, and the most common inciting factor is infection, particularly with HSV. Drug reactions to NSAIDS, anticonvulsants or other drugs play a smaller role. It is of four types, EM minor and EM major, steven jhonson syndrome and toxic epidermal necrolysis. EM generally affects 20-40 years age group, with 20% occurring in children. There may be prodrome of fever, malaise, headache, sore throat, rhinorrhea and cough. The classic skin lesion consist of a central blister or necrosis with concentric rings of variable color around it called typical "target" or "iris" lesion that is pathognomonic of EM.^[1]

CASE REPORT :

A 9 year old girl presented to us with history of ulceration and pain in upper and lower lip since 6 days. She had ulcers in the lower lip and tongue since 6 days. Initially ulcer was present only at the corner of mouth which progressively increased and became severe. Oral lesions appeared first followed by one vesicle at right side of face below lip. Oral lesions were associated with pain which increased on mastication (Figure 1).

Past medical history revealed history of ulceration or cracks at the corner of mouth 1 year back in the winter season. Patient was febrile. Right and left submandibular lymph nodes were palpable and tender. No other relevant medical history elicited. Intraoral examination revealed reduced mouth opening. Swelling of upper and lower lips, fissuring and cracking of right and left corners of mouth with haemorrhagic encrustations was noted on right side of

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Figure 1: Shows lip lesions on the day patient reported. Note the vesicle on cutaneous part of lower lip.

lower lip (Figure 1). It was tender on palpation. Ulceration was also noted on anterior aspect of tongue and vestibular mucosa. Considering history and clinical examination we arrived at a diagnosis of vesiculo bullous disorder. Differential diagnosis included primary herpes simplex infection, Herpes associated erythema multiforme, Pemphigus vulgaris. Primary herpes was ruled out since classical gingival involvement was absent. Pemphigus vulgaris appears as desquamated areas which represent ruptured roof of vesicle or bulla that does not clinically match. Recurrent HSV infection appears as mild disease but our patient had lesions in multiple areas. So a final diagnosis of EM secondary to recurrent HSV infection (HAEM) was made. Patient was advised routine hematological investigations that revealed normal complete hemogram, peripheral smear and erythrocyte sedimentation rate. Serology tests revealed positive blood titre of HSV antigens and four-fold increase in its antibody.

Patient was advised topical triamcinolone gel (0.1%); topical gentian violet (only for external lip); turmeric based mouthwash (0.1%), systemic prednisolone 30 mg was given in three divided doses that was gradually tapered over a period of two weeks. Multivitamin and antipyretics were also advised on first visit. Maintenance therapy of immunomodulator (Levamisole 150mg) was advised thereafter for a period of 3 months.

Patient was recalled after 3 days, where in patient showed scabbing of lower lip lesion, improved mouth opening, healed tongue ulcers. As per patient's mother, lesion had reduced by 50-60%. Patient was recalled after 5 days that showed 80% improvement and 95% improvement after 10 days (Figure 2).



Figure 2: shows significant improvement of lip lesion.

DISCUSSION:

EM is considered to be acute mucocutaneous hypersensitivity disorder. It is characterized by skin eruptions with or without oral or other mucous membrane lesions. It commonly affects young adults of 20-40 years of age and nearly 20% occur in children. It is more common in females than males. In 70% of cases they have a history of preceding herpes simplex infection.^[1] It begins as an acute lesion with or without prodromal symptoms. Patient experiences fever, lymphadenopathy, malaise, headache, cough, sorethroat, polyarthralgia, a week before onset of surface erythema or blisters.^[2] Lesions develop as irregular red macules, papules, vesicles that collapse and gradually increase to form plaques on skin. Skin lesions have a characteristic bulls eye appearance. Oral lesions are referred to as atypical targets. It usually occurs as erythematous macules on lips and buccal mucosa followed by epithelial necrosis, bullae and ulceration with an irregular outline and an erythematous halo. Lip lesions (at vermilion border) are associated with encrustations. EM is of five major types. They are EM minor, EM major, steven Johnson syndrome, toxic epidermal necrolysis and overlap syndrome.^[4] Herpes associated EM (HAEM) is the kind of EM that is caused by preceding HSV infection.

HSV 1 and 2 have known to precipitate EM.^[1] HSV DNA has been found in about 60% of patients clinically diagnosed with recurrent HAEM and 50% of patients with recurrent idiopathic EM by polymerase chain reaction.^[5]

The association between HSV and EM minor was described as early as 1814 by Bateman. Hebra noted it likely occurring in spring and fall when herpetic lesions were common.^[6] Single episode and recurrent EM patients give a history of preceding herpes infection two weeks or less before onset of disease. Acyclovir is said to be very successful in treating such cases since HSV DNA is detected in about 36-81% of cases.^[1]

In a study HSV DNA was found in 60% of cases of single episode or recurrent HAEM and also idiopathic EM. Lip is found to be the most common site of preceding HSV infection in case of recurrent EM. Recent study has also found HSV-1 in 66%, HSV-2 in 28% and both HSV-1 and HSV-2 in 6% of the patients.^[1,7] Half of acute episodes and 80% of recurrent episodes have been associated with HSV.^[8] Antibodies with precipitin reaction to herpes antigen have been identified in sera of 100% of patients with EM and 50% of EM from other causes. This suggests of presence of occult herpes infection or herpes antigen.^[6] Delayed hypersensitivity is considered to be the pathogenesis for HAEM^[9,10] (Figure 3).

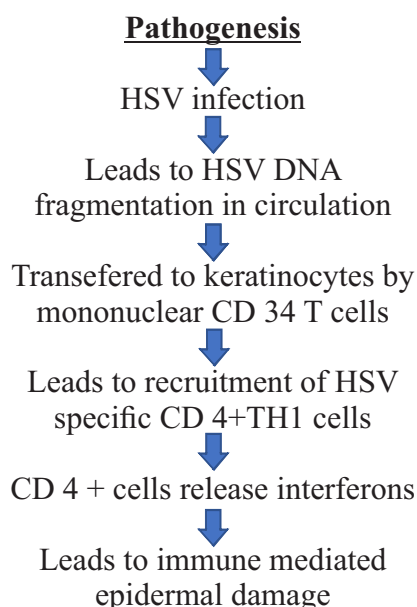


Figure 3: Flow chart elaborating the pathogenesis of HAEM.

HSV genes can be detected by PCR analysis and immunohistochemistry and reverse transcriptase PCR. Previous history of HSV 1 or 2 infection can be

identified by specific IgM and IgG antibodies for HSV 1 or 2. Presence of specific target like lesions on skin makes the diagnosis easy. Mild form of lesions heal in 2-3 weeks although a wide variety of treatment options are available. Mild form of diseases is usually managed by topical antiseptics, topical steroid gels and pain control can be achieved by analgesics. If patient has fever then antipyretics can be used. Topical gentian violet acts as local antibacterial agents thus prevents secondary infection and aids in healing.^[1,3]

Lesions can be recurrent in about 20-25% of cases. Since the disease is also termed as self limiting the lesions usually heal in 10-20 days. HAEM can be effectively managed by systemic acyclovir (200mg, 5 times a day for 5 days) along with antipyretics and analgesics. Systemic steroids are preferred in acute cases along with acyclovir for immediate results.^[11]

Systemic prednisolone 10mg can be given 3 times a day and tapered thereafter. It can also be combined with multivitamins and immunomodulators, especially for maintenance therapy. Mucocutaneous or cutaneous lesions can be treated by topical gentian violet. Since gentian violet has antibacterial properties, it will prevent secondary infection and promote healing.^[11]

CONCLUSION:

Erythema multiforme although is a common disease but effective treatment is very important. Our case is important since our patient is young child who presented with HAEM. She was effectively managed by systemic steroids and gentian violet for the vermilion lesions. Immunomodulators can be used to prevent recurrence in cases of HAEM.

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Conflicts of interest

There are no conflicts of interest.

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