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Case Report

Report of Chronic Erythema Multiforme Major in Pregnancy

Amanda C. Zofkie* and David B. Nelson

Department of Obstetrics and Gynecology, University of Texas Southwestern Medical Center, USA

***Corresponding authors**

Amanda C. Zofkie, Department of Obstetrics and Gynecology, University of Texas Southwestern Medical Center, 5323 Harry Hines Blvd. Dallas, Texas, 75235, USA, Tel: 513-256-3737; Email: azofkie@gmail.com

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Keywords

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- Ulcerative lesions
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Abstract

Background: Erythema mulitforme (EM) is an immune-mediated dermatologic disorder that presents with superficial cutaneous lesions as well as mucosal erosions in varying distributions with widely variant inciting factors.

Case: 19-year-old multiparous African-American female at 12-13 weeks gestation presented with a two-month history of erosive plaques and ulcers on the buccal mucosa, hard palate, and tongue with overlying serosanginous crusting. Following detailed history, serologic laboratory testing, and dermatologic consultation, she was found to have a flare of her chronic EM. Treatment with topical and systemic therapies resulted in resolution of lesions and improved oral intake throughout the remainder of gestation.

Conclusion: Erythema multiforme should be considered in the differential diagnosis of a pregnant patient with ulcerative oral lesions.

ABBREVIATIONS

EM: Erythema Multiforme; HSV: Herpes Simplex Virus; EBV: Ebstein Barr Virus; CMV: Cytomegalovirus; SJS: Stevens-Johnson Syndrome

INTRODUCTION

Erythema mulitforme (EM) is a dermatologic disorder that is immune-mediated and presents with superficial cutaneous lesions as well as mucosal erosions in varying distributions with widely variant inciting factors [1]. Typically, EM is self-limited and a one-time event that may last for 4-6 weeks depending on severity and the presence of mucosal involvement. The most common inciting factor of EM is the herpes simplex virus (HSV). Other inciting factors include other viruses (e.g. Mycoplasma pneumoniae and Ebstein-Barr virus), genetic predisposition, inflammatory bowel disease, medications, and malignancy, whereas other cases are idiopathic. There are also varying types of EM including the minor (no mucosal involvement) and the major (mucosal involvement) subtypes [1]. Despite skin disorders being relatively common in pregnancy, there is limited information regarding the incidence and presentation of EM in pregnancy.

We now present a case of oral ulcerations presenting in the first trimester of pregnancy with subsequent diagnosis of a chronic erythema multiforme major flare. Diagnostic evaluation considerations are discussed, including pregnancy-specific management concerns.

CASE PRESENTATION

A 19-year-old, Gravida 2 Para 1, African-American female at 12-13 weeks gestation presented with decreased oral intake and pain due to a two-month history of ulcerative lesions on her lips. She also noted recent development of pinpoint lesions on her eyelid and intranasal mucosa. On physical exam, she was afebrile and vital signs were within normal limits. Her right eyelid had a small 1 mm dark lesion on the medial epicanthus with several pinpoint 1 mm dark lesions on the bilateral digits and toes. The right nostril had a 1 cm erosive plaque that was scabbed on the nasal septum. The upper and lower lips were grossly inflamed, swollen, cracked and erythematous with overlying yellow flaking with peeling extending to the vermillon borders (Figures 1 & 2). Also shown in the Figure, erosive plaques and ulcers were identified on the buccal mucosa, hard palate and tongue with overlying serosanginous crusting. Her tongue was erythematous with no gross lesions visualized but examination was limited by the patient's inability to fully open her mouth. No genital lesions were identified. Serologic studies identified both Ebstein Barr Virus (EBV) IgG and IgM while cytomegalovirus (CMV) serologies were negative. Prenatal labs were reviewed, all were within normal limits and, of note, and her HIV test was negative. Dermatology services were consulted with recommendation for admission for supportive care, pain control, and further evaluation and management.

Upon further history taking, she reported previous episodes of similar outbreaks in the past. In fact, she reported specifically the diagnosis of erythema multiforme in her father and herself.

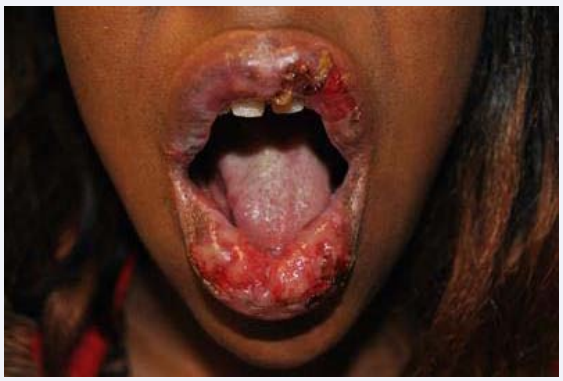

Figure 1 Oral mucosal erosions of EM (coronal view).

Figure 2 Oral mucosal erosions of EM (oblique view).

Prior to pregnancy, she had undergone a biopsy of her oral lesions confirming erythema multiforme major. She had been on several medications including mycophenolate mofetil, oral prednisone, and valacyclovir, all of which she had discontinued at the time of pregnancy recognition. Based upon her history and physical examination findings, she was started on prednisone 60 mg daily for 2 weeks with plan for taper following, topical petroleum ointment to oral lesions, lidocaine oral rinse, and restarted valacyclovir in coordination with dermatology recommendations. Following admission and observation, the patient was discharged on hospital day 3 after she was tolerating adequate oral intake and pain was well controlled with resolution of oral lesions.

DISCUSSION

There are several important features of this case of erythema multiforme in pregnancy, now presented for consideration. First, the differential diagnosis of rash in pregnancy is broad, however, the differential diagnosis for oral lesions is much more narrow and less common-examples of such conditions to be considered include Stevens-Johnson syndrome (SJS), oral herpes virus simplex, pemphigus vulgaris and oral lichen planus as well as EM. Second, exposure to medications in pregnancy should be carefully considered and managed in coordination with an obstetric provider given the teratogenicity risks of certain medications balanced against the benefit of therapy. Lastly, this case highlights the importance of obtaining a detailed medical history, including family history, as this patient had a known history of erythema multiforme major.

Despite being a relatively common condition in reproductive-aged women, there is a paucity of data regarding EM in pregnancy. Very few case reports in the literature detail the disease course or treatment of EM in pregnancy. EM has often been compared and contrasted with Stevens-Johnson syndrome and until 1993-1994 they were previously considered to be part of same spectrum of disease [2]. While currently the gold standard for dermatologic diagnosis is tissue biopsy and histologic evaluation, the pattern of disease, progression of disease, and inciting factors of these two diseases can be quite different. SJS most commonly presents after a drug exposure while EM presents most commonly after infection with the herpes simplex virus. SJS more commonly involves mucosal tissues, while only erythema multiforme major involves mucosa. Knight et al., described 22 pregnant women with SJS in pregnancy in South Africa, all of which were HIV positive. All of these patients presented with SJS after HAART drug initiation [3]. While our patient was HIV negative, the progression of her oral mucosal lesions and distribution was not as aggressive or widespread as the typical presentation of SJS. The patient presented also did not have any new known drug ingestions. While these two diseases can be confused, the patient's family history and personal history of similar episodes assisted in solidifying the diagnosis.

The patient we encountered at our facility is now a contemporary example of EM in pregnancy as well as the first reported case of biopsy-proven chronic EM major in pregnancy. She presented with mucosal oral lesions of the lips and buccal mucosa with resultant inability to tolerate oral intake due to pain. Her case is of interest because her disease was well controlled outside of pregnancy on a strong immunosuppressant (mycophenylate mofetil) and corticosteroid therapy, which was deemed unsafe to continue in the first trimester of pregnancy due to teratogenic side effects. HSV suppression is key to the management of EM, and anti-virals such as acyclovir and valacyclovir are routinely used safely in pregnancy for this purpose. It is still unclear in this case whether the inciting factor for her disease flare was her change in treatment regimen, new onset viral illness, or pregnancy itself. In conclusion, erythema multiforme should be included in the differential diagnosis of oral mucosal lesions in the pregnant patient.

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