Published in final edited form as:
Arch Dermatol. 2009 August ; 145(8): 952–953. doi:10.1001/archdermatol.2009.159.

Pregnancy-associated dermatomyositis

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Dermatomyositis (DM) is an idiopathic inflammatory myopathy with characteristic skin manifestations. Reports of the association between DM and pregnancy are rare, though two types of pregnancy-related DM have been proposed: one presenting during pregnancy and the other less common type developing postpartum.[1] Three reported cases of the latter exist including a 33-year-old woman who manifested symptoms of classic DM (CDM) five days after her first delivery, a 29-year-old woman who developed DM one month after a normal delivery and a woman who was diagnosed fifteen days after a cesarean section.[1,2,3] In addition, two cases describe postpartum exacerbation in which a 31-year-old woman who experienced periungual erythema at 32 weeks gestation developed full-blown CDM three months after delivery and another woman who suffered a flare of her childhood DM during the post-spontaneous abortion period.[3,4] Various triggers for pregnancy-associated DM may include exposure of the mother to fetal antigens and maternal hormonal changes.[1] We report a patient with amyopathic DM (ADM) that developed after a spontaneous abortion and progressed two years later to CDM after the delivery of a healthy infant.

A 38-year-old woman was diagnosed with ADM based on clinically and histologically typical skin findings beginning four days after a spontaneous abortion. Physical examination revealed forehead and malar erythema with scale, gottron's papules over the bony prominences of her hands and elbows and mild cuticular telangiectasias. A skin biopsy was compatible with DM (Fig 1). She denied any muscle weakness or dysphagia. Further testing revealed negative antinuclear and anti-Jo1 antibodies, normal aldolase and creatine kinase (CK) levels, as well as normal electromyography (EMG) and pulmonary function testing (PFT). A malignancy work-up was unremarkable. A three-week course of oral prednisone significantly improved her inflammatory skin findings and hydroxychloroquine stabilized her disease activity.

Seven months later, the patient achieved pregnancy with in vitro fertilization. Her ADM remained stable throughout this period. However, four months after a normal delivery, the patient's skin gradually worsened and she developed pruritic erythematous plaques on her face, v-neck erythema and gottron's papules on her hands and elbows (Fig 2). After three months, she also experienced difficulty climbing stairs and lifting her daughter. Muscle enzymes were elevated with a CK level of 8,241 U/L (ref ≤ 165) and aldolase level of 53.6

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Author Contributions: Mina Yassaee and Drs. Victoria P. Werth and Carrie Kovarik had full access to all of the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. *Study concept and design:* Yassaee and Werth. *Acquisition of data:* Yassaee, Kovarik, Werth. *Analysis and interpretation of the data:* Yassaee, Kovarik, Werth. *Drafting of the manuscript:* Yassaee, Kovarik, Werth. Critical revision of the manuscript for important intellectual content: Kovarik, Werth. *Statistical analysis:* not applicable. *Obtained funding:* not applicable. *Administrative, technical or material support:* Yassaee, Kovarik, Werth. *Study supervision:* Werth.

Financial Disclosure: None reported.

U/L (ref ≤ 8.1) and an EMG confirmed the presence of myositis. Treatment with high-dose prednisone and azathioprine improved her muscle weakness and normalized her enzymes after two months.

Of note, the patient started the first of two cycles with clomiphene citrate the day her rash first appeared. In hope of a second pregnancy, she received one more treatment a month prior to her muscle flare. Ovarian hyperstimulation following six to ten cycles of therapy has been associated with the induction or exacerbation of systemic lupus erythematosus in women[5]. The timing and number of cycles make clomiphene a less likely trigger of DM in this case. However, hormonal fluctuations potentially play an important role in the pathogenesis of DM.

Acknowledgments

None.

Funding/Support: Not applicable.

Role of the Sponsors: Not applicable.

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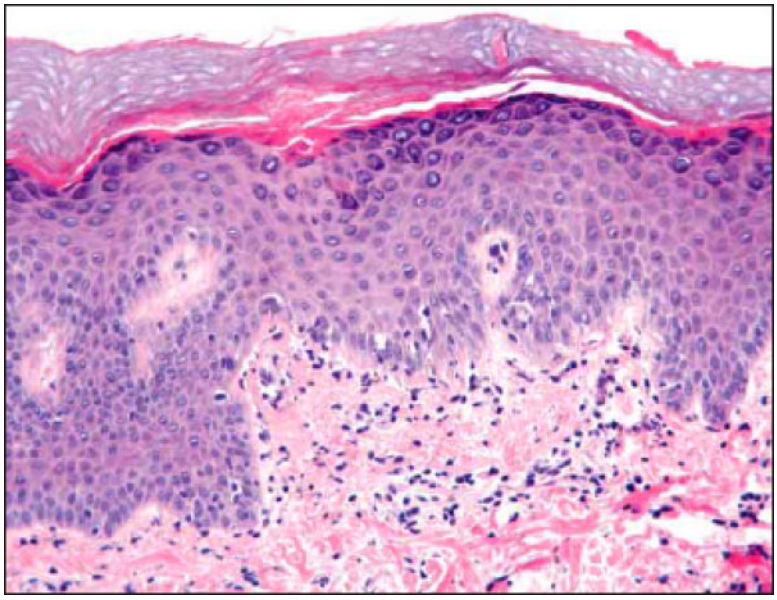

Figure 1.
Skin biopsy of right middle finger, demonstrating an interface dermatitis, characterized by a patchy band-like lymphocytic infiltrate in the superficial dermis, basal vacuolar alteration of the epidermis and necrotic keratinocytes. (Hematoxylin-eosin stain x 100).

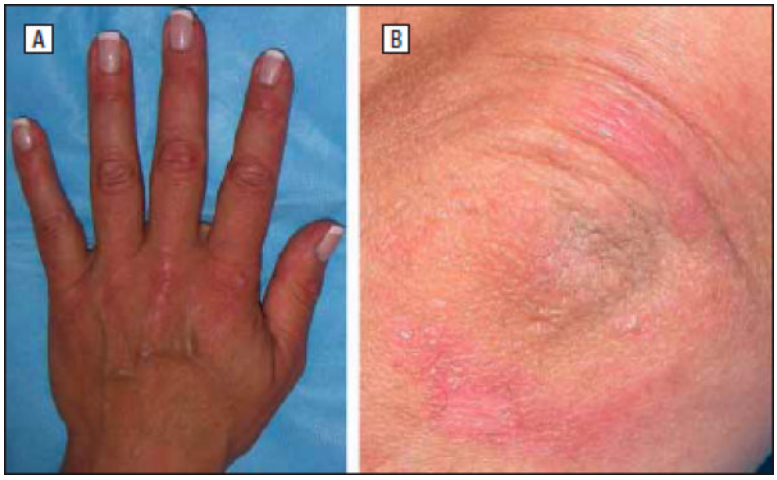

Figure 2.
Gottron's papules on the knuckles (A) and elbow (B).