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Lyell syndrome in pregnancy: A case report

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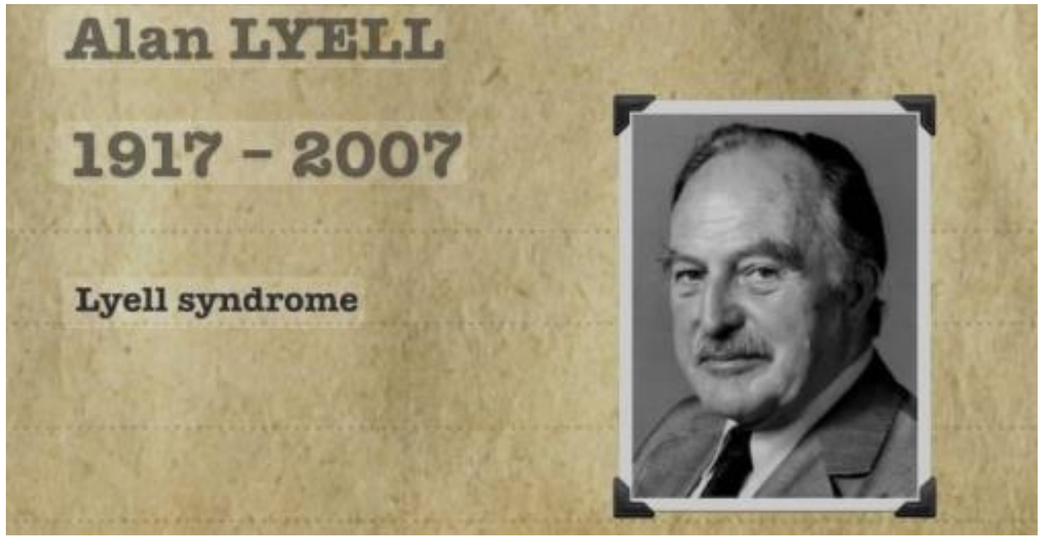

Introduction

Lyell syndrome or Toxic Epidermal Necrolysis (TEN) is a serious drug reaction characterized by extensive epidermal necrosis and rapid expansion. The culprit drugs implicated in most series are antibacterial sulfonamides, anticonvulsants, allopurinol, pyrazolone derivatives and less frequently, other nonsteroidal anti-inflammatory drugs. Rare cases of paracetamol-induced Lyell syndrome have been also described, but never in pregnancy after paracetamol administration, neither in association with midodrine treatment.

Case report

We present a 36 year-old pregnant woman (gravida 2, para 1) at the 7th week of gestation was admitted to the Intensive Care Department for a paracetamol or midodrine-induced Lyell syndrome. Her medication history only consisted of a single use of acetaminophen 1000 mg and daily therapy with midodrine 2,5 mg for hypotension.

Results

Twelve hours later patient the last drug assumption, the patient reported some initial signs of skin toxicity, starting with some erythema and blistering on the face and back and she was admitted to the hospital. The symptoms extended to the trunk and the upper thighs during the next 2 days; systemic corticoid therapy (methylprednisolone 40 mg), chlorphenamine 10 mg and pantoprazole 40 mg were initiated. Moreover, cetirizine 10 mg was administered orally. The skin lesions rapidly worsened with profound erythema and blisters on the neck, thorax, and arms covering over 70% of her body surface area, and purulent discharge from the eyes and daptomycin 4 mg/kg/die was started. Following this rapid progression of skin toxicity, the patient was transferred to the intensive burn trauma center. Her general condition deteriorated further more requiring a systemic antibiotic medication with amoxicillin/clavulanic acid 2000 mg/200 mg every 12 hours and human immunoglobulins 0,4 g/kg for 5 days. A significant decreas of hemoglobin and hematocrit levels was recorded and ferrous sulphate 256,3 mg/day and folic acid 5 mg/day treatment were added. Histologic diagnosis revealed epidermal necrosis and interface dermatitis along the dermoepidermal junction zone compatible with Lyell syndrome. The patient was discharged twenty days after the admission with scar lesions covering over 70% of her body surface area. The delivery was uneventful, and the healthy female infant (weight: 2940 g, Apgar score: 9/9/9) had no signs of TEN or sequelae resulting from the therapy for the mother.

Conclusions

A combination of genetic susceptibility and an altered immune system during pregnancy may contribute to the pathogenesis of TEN, involving a cytotoxic T-cell mediated reaction with release of inflammatory cytokines. Mother-to-fetus transmission of TEN is rare. Due to high risk of mortality, management of patients with TEN requires rapid diagnosis, identification and interruption of the culprit drug. Intensive care unit admission, supportive treatment and high-dose intravenous immunoglobin therapy and systemic corticosteroids are required. Moreover, this pharmacological approach does not need to be modified in pregnant patients.

Treatment