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RECURRENT IMPETIGO HERPETIFORMIS TREATED SUCCESSFULLY WITH CYCLOSPORINE: A CASE REPORT

Dermatology

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ABSTRACT

Impetigo herpetiformis (IH) is a rare dermatoses with potential serious consequences for both the mother and fetus. Treatment is difficult and steroids is the mainstay of treatment. Cyclosporine has been used for a few cases resistant to steroids. We report our own experience in case of IH of 21 year-old woman treated successfully with tapering doses of oral prednisolone and cyclosporine in her two successive pregnancy. The disease was refractory to the oral steroids, therefore, treatment with cyclosporine was initiated and a rapid regression of the lesions was observed. Gestation was maintained, with a good perinatal outcome. Both the fetus and mother monitored closely when systemic illness occurs, as there is a risk of stillbirth. Cyclosporine, when used appropriately is effective and relatively safe.

KEYWORDS:

impetigo herpetiformis, cyclosporine

INTRODUCTION

Impetigo herpetiformis is a rare pustular form of psoriasis occurring in pregnancy. IH having tendency to be symmetrical and grouped, often starting in the flexures, and the constitutional disturbances may be severe. [1-4]

The onset is usually in the last trimester of pregnancy, but may be earlier and has been recorded in the first month of pregnancy. [2]The disease tends to persist until the child is born and occasionally even later. We report here a case of pustular psoriasis in her two succesive pregnancy, which was successfully treated with tapering doses of oral prednisolone and cyclosporine.

CASE REPORT

A 21year-old primigravida of 32 weeks gestation presented with sudden onset crops of pustular lesions and associated fever and chills. Lesions started as pustules on the thighs,abdomen, chest and scalp which then became generalized. Cutaneous examination revealed multiple grouped pustules and over few places coalescing to form "lakes of pus"[figure1]

Laboratory investigations includes complete blood count, liver function tests, renal function tests, blood sugars, urine, stool examination were within normal limits. Her VDRL, enzyme-linked immunosorbent assay for human immunodeficiency virus, serum electrolytes, serum calcium and phosphorus were also normal. Pustules were sterile and histopathology showed characteristic features of pustular psoriasis. A diagnosis of pustular psoriasis of pregnancy was made and the patient was started on oral prednisolone 40 mg daily. Patient had full term normal delivery at 36 weeks of gestation. Skin biopsy was done post-delivery which was suggestive of impetigo herpetiformis. The lesion resolved post delivery. After one month patient again developed new lesion over thighs. Patient was started on tablet prednisolone 40 mg OD, since there was partial improvement, the decision was taken to add oral cyclosporine after consulting obstertrician and paediatrician at the dose of 50 mg bid (2 mg/kg/day) and gradually tapering(10 mg /fortnightly) oral prednisolone. We had continued oral cyclosporine for three months postpartum and then stop as lesion resolved completely.

After two years, same case was referred by the attending physician of private hospital to our hospital, 37 weeks gestation and with pustular skin lesions. As patient responded very well to oral prednisolone and cyclosporine in previous episode, after consulting obstetrician and paediatrician, we again started systemic steroids (40 mg) and cyclosporine 50mg bid (2mg/kg). Vaginal delivery of a healthy child was done at 38 weeks of gestation in private hospital. Skin lesions improved progressively and rapidly during postpartum and steroid dosage were gradually tapered (10 mg every fortnightly). Oral cyclosporine was continued for three months for complete remission of disease. There was no evidence of new lesion after four months of postpartum period in follow up.

DISCUSSION

Pustular psoriasis of pregnancy is characterized by an acute eruption of erythematous patches studded with pustules at the margins, occurring usually in the last trimester of pregnancy and has been recorded in the first month of pregnancy. [2]

The lesions originate at the flexures, then spread to the center and may become generalized; occasionally there may be painful oral erosions and involvement of subungual areas of nails leading to onycholysis. The face, palms, and soles are commonly spared. The rashes may be pruritic or painful, accompanied by constitutional symptoms of fever, chills, malaise, diarrhea, nausea, and arthralgia. Rarely tetany, delirium, and convulsions can occur, if the hypocalcemia is severe. The disease tends to persist until the child is born and occasionally long afterwards.[2] Recurrence has been described in up to nine pregnancies, and on subsequent use of oral contraceptive.[2,3] Laboratory derangement includes leucocytosis, neutrophilia, elevated ESR, anemia, and hypoalbuminemia. Hypocalcemia has often been reported. [1, 2] Differential diagnosis includes pemphigoid gestationis, pustular drug eruption and subcorneal pustular dermatosis. Pemphigoid gestationis usually occurs at the first week of pregnancy and may flare up immediately postpartum, in contrast to impetigo herpetiformis. Moreover, the initial lesions consist of pruritic urticarial papules and plaques, target lesions, and annular wheals. Pustular drug eruption will have a definite drug-intake history. Subcorneal pustular dermatosis lesions also have flexural and trunk predilection, but the lesions are pustular from the beginning, flaccid, turbid, and often oval rather than circular. Some lesions show a level of pus with clear fluid above.

In our patient, the onset of pustular eruption was at the third trimester of pregnancy, in the flexures, with marginal pustules becoming generalized later. The rashes were accompanied with constitutional

symptoms of fever, chills, malaise, and burning pain over the lesional skin. Laboratory findings were within normal limits, with pus cultures being negative.

Pustular psoriasis of pregnancy is very rare and by 1982, only 200 cases have been documented. [2]. In more severe and longstanding disease, greater is the risk of placental insufficiency, leading to stillbirth, neonatal death, or fetal abnormalities [3, 5, 6, 10].

Oral corticosteroid is the mainstay of treatment in the management of pustular psoriasis of pregnancy. Of late, cyclosporine as well as methotrexate have been used in this condition. [7, 8, 9]. Cyclosporine, which is categorized as pregnancy category 'C', has been successfully used at doses between 2-5 mg/kg weight daily, to treat cases that are refractory to a high dose of systemic corticosteroids. Cyclosporine is a natural cyclic peptide compound of 11 amino acids, metabolized in the liver by P450, 34A cytochrome, and excreted primarily by way of bile, through feces. Only 6% of the total dose is excreted in urine and minimally through breast milk.

CONCLUSION : The drug of choice for IH is oral steroids (prednisolone). However in our case of impetigo herpetiformis, which was unresponsive to oral corticosteroids,cyclosporin found to be effective without any adverse effect to both mother and fetus. The drug was tapered and withdrawn over a short period after delivery, to avoid possible side effects.

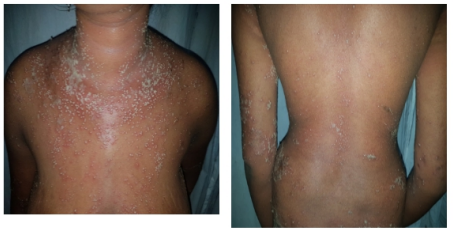

Figure 1,2 showing multiple discrete and confluent pustules present over chest, back and bilateral upper arm

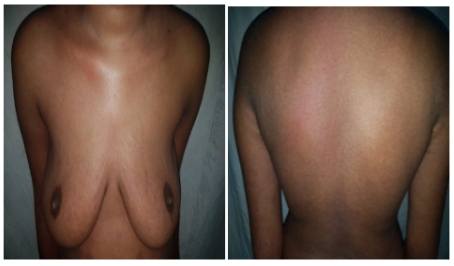

Figure 3,4, showing complete resolution of lesion after cyclosporine therapy

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