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Toxic Epidermal Necrolysis Successfully Salvaged with Cyclosporine

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ABSTRACT

Stevens-Johnson syndrome (SJS) and toxic epidermal necrolysis (TEN) are severe, life-threatening mucocutaneous disorder, with the adult incidence being estimated to be 0.4-1.2 cases/million; most commonly caused by drugs, with a high morbidity and mortality that require immediate medical care. I hereby report a case of SJS treated successfully with cyclosporine.

Keywords: Stevens-Johnson syndrome, Toxic epidermal necrolysis, Cyclosporine

INTRODUCTION

Stevens-Johnson syndrome (SJS) and toxic epidermal necrolysis (TEN) are severe, life-threatening mucocutaneous disorder, with the adult incidence being estimated to be 0.4-1.2 cases/million; most commonly caused by drugs, with a high morbidity and mortality that require immediate medical care [1]. Systemic corticosteroids have been utilized in the management of SJS/TEN, but their use is still not universally accepted [2]. Cyclosporine, intravenous gammaglobulin, cyclophosphamide, thalidomide plasmapheresis has been used in its treatment with varying success [1]. I hereby report a case of SJS treated successfully with cyclosporine.

CASE PRESENTATION

A 59 year old male, a known diabetic for 10 years, presented to us with painful, erythematous, non-itchy rash over the face, back, buttocks and genital region for 4 days, who subsequently developed fluid filled lesions and erosions, redness and watering from the eyes, oral and genital ulcers within 24 h (Figure 1). The eruption followed intake of paracetamol and diclofenac combination for pain in the legs. On examination there were necrotic areas with sheets of epidermal detachment over the face, back and buttocks; ocular examination revealed mucopurulent conjunctivitis; genital region revealed confluent ulcers involving the glans, shaft of penis and scrotum. Total body surface area involved was 35%. Nikolsky's sign was positive. He was immediately admitted. His heart rate was 92 bpm, blood urea, 22 mg/dl, serum glucose, 283 mg/dl, serum bicarbonate, 24 mmol/L, haemoglobin 12.4 g/dl, total leukocyte count, 6300/cmm and total platelet count 1.9 lac/cumm. There was no evidence of malignancy. A final diagnosis of toxic epidermal necrolysis with a scorten of 3 was made. Apart from the supportive care he was started on oralcyclosporine 100 mg twice daily

(3 mg/kg/day). The patient showed marked improvement within 10 days of treatment (**Figure 2**).

DISCUSSION

SJS eponymously named after Dr. Albert Mason Stevens and Dr. Frank Chambliss Johnson, who in 1922 first described cutaneous eruptions, severe ocular and oral involvement in two young boys [3]. In 1956, Lyell introduced the term toxic epidermal necrolysis [1]. In 1983, based on their similar histolopathologic findings, SJS and TEN were synonymously associated with erythema multiforme major but later Bastuji-Garin et al. in 1993 and Roujeau in 1994 proposed the differentiation of erythema multiforme from SJS and TEN based on clinical and etiologic information [3].

Majority of cases are drug induced, high risk drugs being phenytoin, carbamazepine, lamotrigine, olanzapine, ofloxacin. non-steroidal anti-inflammatory drugs, sulphonamide antibiotics and nevirapine; [1,3] while some cases have been said to be triggered by infections the most common being mycoplasma pneumonia [4]. The disease involves extensive necrosis of epithelial keratinocytes of the skin and mucous membrane caused by cytotoxic Tlymphocytes with the associated triggers being soluble as ligand, perforin and granzyme [4].

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Figure 1. a) Multiple erosions on the face especially the periocular and perioral region with watering from eyes; b and c) multiple erosions over the back showing cigarette paper thinning at the margins; d) well defined erosions over both the buttocks; e) extensive erosions on the penis as well as scrotum.



Figure 2. a-c) Marked re-epithelisation seen all over after 10 days of therapy; d) Drying of the lesion in the genital region.

CONCLUSION

Cyclosporine, a calcineurin inhibitor, is frequently used in the treatment of organ transplant and dermatologic conditions like psoriasis and atopic dermatitis. In SJS-TEN it causes inhibition of inflammatory cytokines released due to activation of cytotoxic T-cells and reduces keratinocyte apoptosis [5]. We present the above case to display the effectiveness of cyclosporine alone without using corticosteroid in the management of TEN.

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