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Generalized bullous fixed pigmented erythema mimicking toxic epidermal necrolysis: About two-cases

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Introduction:

Fixed pigmented erythema (FPE) is a cutaneous adverse drug reaction characterized by recurrent skin lesions that reappear at identical sites upon re-exposure to the causative agent. The generalized bullous form is rare and may clinically mimic toxic epidermal necrolysis (TEN). This resemblance can lead to inappropriate management, highlighting the importance of accurate diagnosis.

We report two cases of generalized bullous FPE initially suspected to be Lyell syndrome, where clinical evolution and histopathological findings enabled correct diagnosis and appropriate therapeutic management.

Case 1: A 58-year-old woman with no significant medical history developed a generalized pruritic bullous eruption 24 hours after taking etoricoxib. On examination, she was hemodynamically and respiratorily stable, with erythematous macules on the trunk and limbs, extensive post-bullous erosions, a positive Nikolsky sign, with a “wet sheet” appearance on the back, and fissured cheilitis. Laboratory tests were unremarkable. Skin biopsy revealed minimal epidermal necrosis with perivascular lymphocytic infiltration. Drug causality assessment implicated etoricoxib. The patient improved with dual antihistamine therapy and local wound care.

Case 2:

A 51-year-old man, with a history of schizophrenia treated with levomepromazine and diazepam for 10 years, presented with generalized erosions that developed within 24 hours after taking omeprazole. On admission, the patient was conscious, hemodynamically and respiratorily stable, and exhibited large post-bullous erosions on the back, buttocks, and thighs, along with hyperpigmented macules on the trunk and a positive Nikolsky sign, but no oral or conjunctival mucosal involvement. Laboratory tests were unremarkable, and the biopsy supported a diagnosis of FPE. The pharmacological investigation implicated omeprazole as the cause. The patient’s condition improved favorably with antihistamines and topical ointment dressings.

Conclusion

Generalized bullous FPE although rare, poses a major diagnostic challenge due to its clinical similarity to TEN. The distinctive feature of these cases lies in the absence of severe systemic involvement despite extensive skin lesions, along with a misleading positive Nikolsky sign, and favorable progression with simple antihistamine treatment eliminating the need for aggressive therapy or intensive care measures typically reserved for Lyell syndrome. These cases highlight the importance of histology and pharmacological investigation to avoid inappropriate management but performing a skin patch test that was not possible in these cases remains the key to diagnosis. The drugs most frequently implicated in FPE include nonsteroidal anti-inflammatory drugs (such as etoricoxib), antibiotics (sulfonamides, tetracyclines), anticonvulsants, and, more rarely, proton pump inhibitors (omeprazole). This warrants heightened vigilance when prescribing these medications, especially in cases of recurrent lesions at the same sites**.**