INFLAMMATORY SKIN DISEASES (OTHER THAN ATOPIC DERMATITIS & PSORIASIS)

A CASE OF PG IN A PATIENT WITH UNTYPICAL SAPHO SYNDROME

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Background: Pyoderma gangrenosum (PG) is a rare inflammatory neutrophilic dermatosis manifesting as painful ulcers with violaceous, undermined borders on the lower extremities. SAPHO (synovitis, acne, pustulosis, hyperostosis, osteitis) syndrome manifests in several skin disorders including palmoplantar pustulosis, psoriasis pustulosa and acne conglobata. However, PG in a patient with SAPHO syndrome is extremely rare. Herewith we present a detailed description of a case of PG in a patient with untypical SAPHO syndrome.

Observation: A 56-year-old woman presented with a 3-year history of erythematous plaques on her right lower leg, with extensive ulceration for 1 year. The patient has received skin grafting since the ulceration with striking redness, increased pain and oozing significantly affected her life. However, erythematous plaques and ulceration with great pain recurred around the post-transplant skin lesion. Besides, her left arm was amputated 1 year before because of severe left elbow osteomyelitis with a 16-year history of aggravated left elbow joint pain. Inflammatory bowel disease, other related diseases and family history of PG or arthritis were denied. After admitted , a skin biopsy was toke from the plaque on the right lower leg, revealing dense infiltration of mixed inflammatory cells in the dermis. Radionuclide bone scanning revealed intense activity at sternal stem, sternoclavicular joints and proximal clavicle, presenting a so-called "bull's head" change. After intravenous administration of methylprednisolone 40mg per day, the skin lesion stopped distending and the pain released. Besides, there is a key clue that the parents of the patient were cousins. So the work to mine some gene mutations associated with neutrophilic dermatoses is underway.

Key message: To date, there has been few reported cases of PG presenting as a skin manifestation of SAPHO syndrome. This case is a rare and unusual case of neutrophilic dermatoses, and highlights the association between SAPHO syndrome and PG.