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PHOTOLETTER TO THE EDITOR

Oral ulceration in pyoderma gangrenosum

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Abstract

A 65-year-old woman presented with widespread necrotising cutaneous ulceration and oral involvement. Past history included rheumatoid arthritis, and a left nephrectomy.

Examination revealed multiple violaceous undermined ulcers. Blood investigations showed an acute inflammatory response. Skin histopathology showed epidermal ulceration with acute and chronic inflammation. Direct immunofluorescence was negative. A diagnosis of pyoderma gangrenosum with oral involvement was made. Mycophenolate mofetil therapy resulted in complete resolution of her pyoderma gangrenosum. Her treatment was complicated by a left proteus mirabilis psoas abscess. This resolved following four weeks of antibiotics.

Pyoderma gangrenosum with oral involvement is rare but has been linked with inflammatory bowel disease and hematological disorders. Oral pyoderma gangrenosum has not previously been described in rheumatoid arthritis. Primary psoas abscess is rare but can develop in immunocompromised patients. Proteus mirabilis has been reported in patients years after nephrectomy. This is a rare case of pyoderma gangrenosum with oral involvement. (*J Dermatol Case Rep*. 2011; 5(2): 34-35.)

Key words:

mycophenolate mofetil, oral mucosa, oral ulceration, psoas abscess, pyoderma gangrenosum

A 65-year-old woman presented with a six week history of widespread necrotising cutaneous ulceration. She had a prodrome of superficial ulceration on the lower abdomen that had healed spontaneously. Four weeks later she developed pustules that rapidly progressed to intensely painful deep ulceration involving the tongue, both breasts, oral mucosa, abdomen, perianal skin and feet. Past medical history included rheumatoid arthritis which was quiescent, a left nephrectomy in 1974 for nephrolithiasis and osteoarthritis of the right knee. She smoked 20 cigarettes a day. Her medications were occasional mefenamic acid and paracetamol.

Clinical examination revealed multiple violaceous undermined areas of ulceration measuring 8-12 cm in diameter on the above sites (Fig. 1). There was recent scarring on the lower abdomen. The buccal mucosa and tongue were ulcerated with a deep 3 cm ulcer on the lateral border of the tongue (Fig. 2). Fresh pustules were present on the abdomen and arms. Systemic vasculitis or atypical pyoderma gangrensoum was suspected.

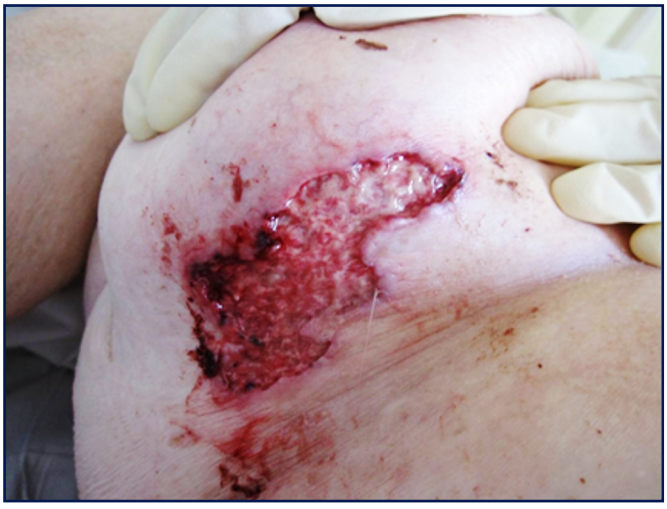

Figure 1
Typical pyoderma gangrenosum ulcer with violaceous undermined borders.

Haematological and biochemical indices were normal except for a leucocytosis (14.86 x109/l) with a neutrophilia (10.62 x109/l), a raised C-reactive protein (77 mg/L) and erythrocyte sedimentation rate (82 mm/hr). Histopathology of the skin showed epidermal ulceration with intense acute and chronic inflammation and no vasculitis. Direct immunofluorescence was negative, as were ZN stain and fungal cultures. A vasculitis screen showed a positive antinuclear antibody with a titre of 1:400, rheumatoid factor of 252 with anti-cyclic citrullinated peptide antibody 32 units/mL. Dipstick urinalysis demonstrated persistent hematuria and proteinuria with no casts. Renal biopsy of the solitary kidney was normal with no vasculitis. A diagnosis of pyoderma gangrenosum (PG) with oral involvement was made.

Pulsed intravenous methylprednisolone 1 gram, broad spectrum antibiotics and oral prednisolone 60 mgs/day resulted in dramatic reduction in pain within 48 hours. Mycophenolate mofetil (MMF) 2 grams/day was then introduced. Her mucosal and cutaneous ulcers healed over 5 weeks. Three weeks after commencing treatment, she developed left flank pain and a palpable swelling. Computerised tomography confirmed a psoas abscess which drained 600 mls of purulent fluid and cultured Proteus mirabilis. This resolved following two weeks of intravenous tazobactam/piperacillin followed by two weeks of oral ampicillin. Four weeks later after erroneously discontinuing her MMF, she represented with recurrent oral ulceration, right flank pain, pyrexia, nausea and dysuria. CT and urine culture confirmed a Escherichia coli pyelonephritis which resolved with two weeks of intravenous aztreonam.

Reinstituting MMF resulted in complete resolution of PG in this patient and allowed discontinuation of corticosteroids. MMF was preferred to Cyclosporin A because of the solitary kidney. A retrospective review of MMF in PG showed over half of patients gain complete ulcer resolution.[1]

Pyoderma gangrenosum with oral involvement is rare. There have been a handful of case reports in the literature linking oral PG with inflammatory bowel disease and hematological disorders.[2] Oral PG in the absence of underlying systemic disease has also been described.[3] To date, oral involvement in PG has not been described in the setting of rheumatoid arthritis. Primary psoas abscess is also rare but can develop in immunocompromised patients.[4] The most frequent pathogen is Staphylococcus Aureus, however Proteus has been reported in patients years after nephrectomy and with gastrointestinal disease.[5]

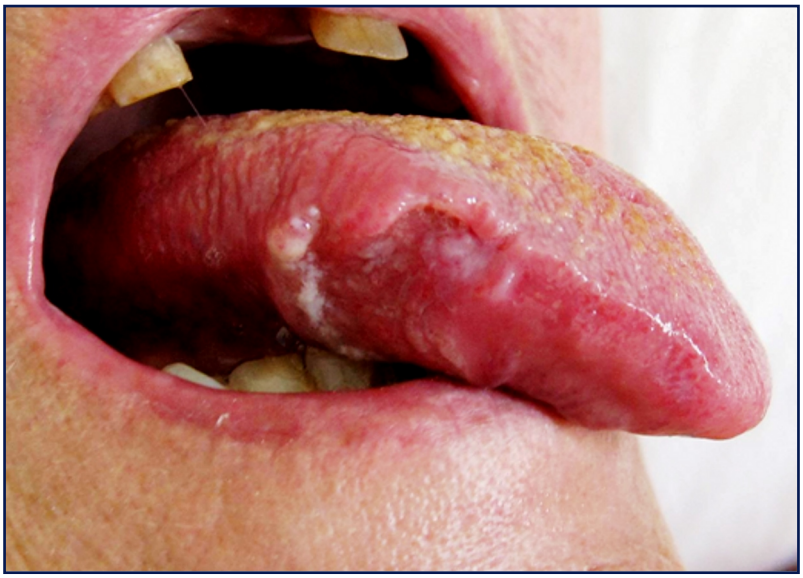

Figure 2
Deep ulceration on lateral tongue.

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