SHORT COMMUNICATION

Recurrent Neutrophilic Dermatosis of the Face Successfully Treated with Potassium Iodide

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Recurrent neutrophilic dermatosis of the face (RNDF) is a rare disorder of neutrophilic dermatosis (ND), characterized by recurrent, painful erythematous plaques confined to the face, with a sterile neutrophilic infiltrate and no vasculitis on histopathology (1–5). We present a case of recurrent neutrophilic dermatosis affecting not only the face but also the head and neck, which was successfully treated with potassium iodide.

CASE REPORT

A 48-year-old Japanese male presented with a 1-year history of a painful rash on the face, head, and neck, without general symptoms such as malaise, fever, and arthralgia. Initial treatment at another hospital with topical steroids and oral antihistamines slightly alleviated the lesions, but they recurred repeatedly. He had an 18-year history of hypertension, treated with benidipine hydrochloride. The patient denied any triggering events, such as trauma and prolonged sun exposure, before the development of the lesions. Physical examination revealed multiple tender, painful erythematous plaques and nodules on the face, head, and neck, along with erythema and painful swelling of the upper eyelids (**Fig. 1**A–C). The lesions were limited to these regions. Blood tests showed leukocytosis of 10,590/μL (normal; 3,300–8,600/μL) with 67.9% neutrophils and an elevated C-reactive protein level of 0.90 mg/dL (normal; <0.30 mg/dL); other values showed no abnormalities, including rheumatoid factor, antinuclear antibodies, and soluble interleukin-2 receptor. Based on these findings, the differential diagnosis included sarcoidosis, malignant lymphoma, and Hansen's disease. A skin biopsy of the lateral upper eyelid erythema revealed marked neutrophil infiltration with some lymphocytes and histiocytes in the dermis and subcutaneous tissue (**Fig. 2**A, B). The other skin biopsy of the erythematous plaque on the neck revealed an infiltration of multiple neutrophils and some histiocytes in the dermis (Fig. 2C, D). Neither skin biopsy specimen showed evidence of vasculitis or fibrinoid necrosis, and no pathogens were detected by periodic acid-Schiff or Grocott staining. Therefore, a diagnosis of ND was made. The patient was treated with potassium iodide at 0.9 g/day without topical steroids, and the lesions completely resolved within 1 month (Fig. 1D). After 10 months of treatment, erythema and painful swelling appeared on the right upper eyelid, without any triggering events or the introduction of new medication. The dose was increased to 1.5 g/day, which improved the lesion within 1 week. Over 5 years on 0.9 g/day, recurrence occurred only twice.

DISCUSSION

ND is an inflammatory skin disorder characterized by a sterile, predominantly neutrophilic infiltrate on histopathology and includes Sweet's syndrome (SS), pyoderma gangrenosum, Behçet's disease, and other conditions (6). RNDF, an uncommon form of ND, was first described by Whittle et al. in 1968 in 2 patients with recurrent tender erythematous plaques on the face (1). Although previously reported patients with RNDF were compatible with a histopathological diagnosis of SS, no general symptoms or associated conditions were observed (1–5). Additionally, cases of ND localized to other sites, such as the hands, have been proposed as a subtype of SS (7). However, controversy surrounds whether these cases, including RNDF, represent subtypes of SS or independent entities.

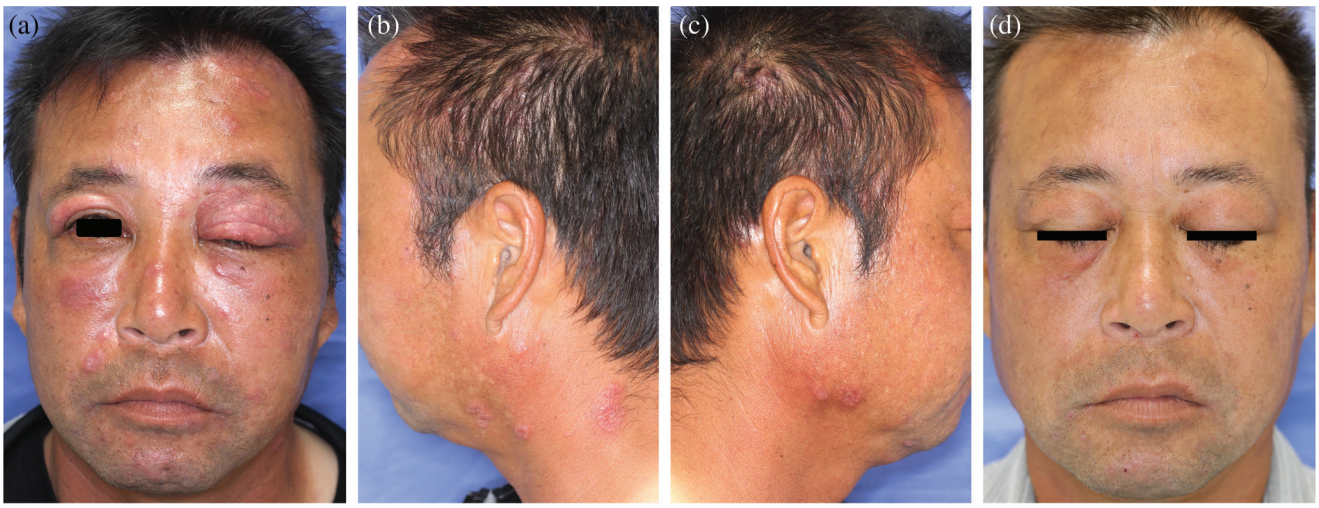

Fig. 1. Clinical findings. (A–C) Clinical images of multiple tender, painful erythematous plaques and nodules on the face, head, and neck, and erythema and painful swelling of the upper eyelids were noted. (D) These lesions resolved completely within 1 month.

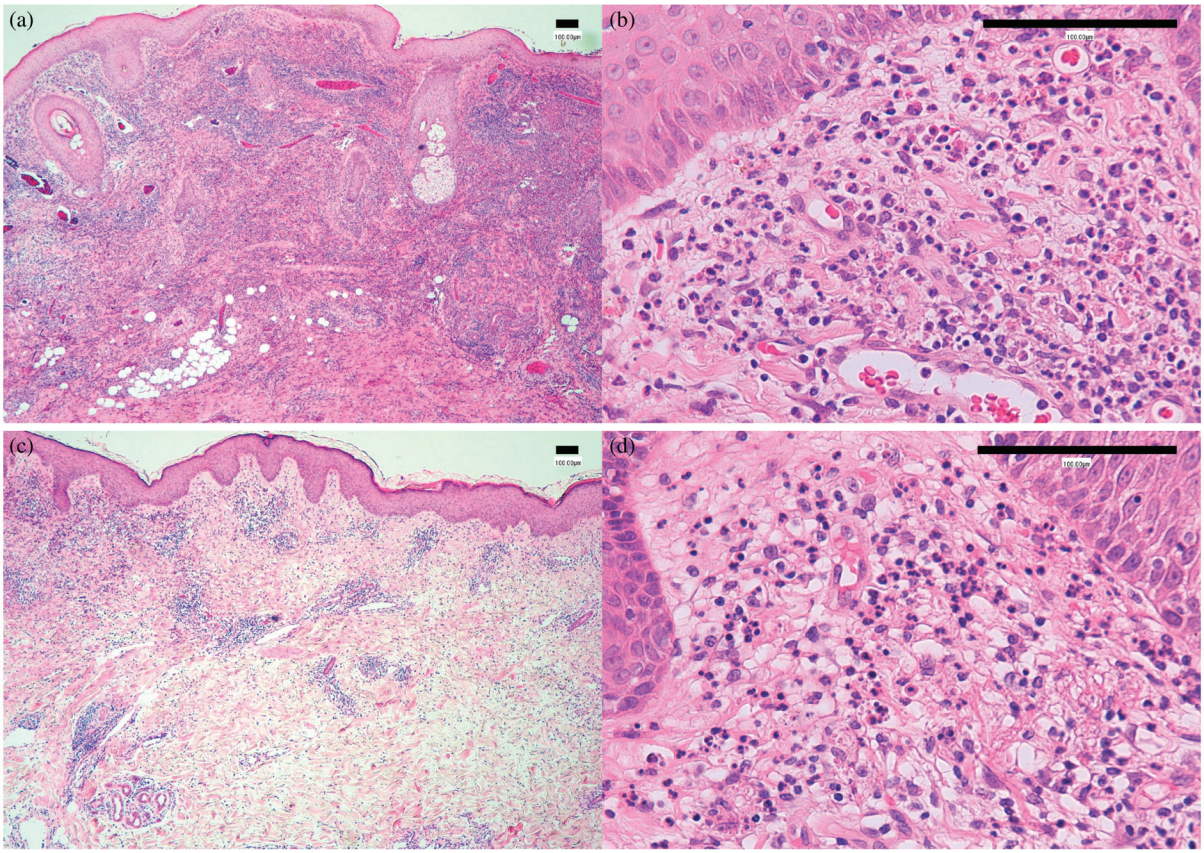

Fig. 2. Histopathological findings. (A, B) A haematoxylin-eosin-stained slide of the lateral upper eyelid erythema showed marked neutrophil infiltration with some lymphocytes and histiocytes in the dermis and subcutaneous tissue. In addition, blood clots were seen in the dilated blood vessels in the dermis (bars in the image = 100 μm). (C, D) The other skin biopsy from the erythematous plaque on the neck revealed dilated blood vessels and an infiltration of multiple neutrophils and some histiocytes in the dermis (bars in the image = 100 μm).

To date, 8 patients with RNDF, including the current case, have been reported in the literature (1–5). RNDF predominantly affected females (75%), and the age ranged from 23 to 57 years (mean: 40 years). All patients presented with recurrent tender erythematous plaques and in 1 patient developed into vegetative crusted plaques (5). The affected areas in our patient are unique because the lesions involved not only the face but also the head and neck, which has not been reported in the literature. This suggests that skin lesions of RNDF rarely develop on the head and neck.

Fever was observed in only 1 patient (13%), and leukocytosis was found in 3 patients (38%) (1). Moreover, none of the cases had associated conditions such as infection, haematological malignancy, or inflammatory bowel disease. In the current case, during the 5-year follow-up period, neither general symptoms associated with RNDF nor associated conditions were observed.

The primary treatment for ND is prednisolone (PSL), with dapsone, colchicine, and potassium iodide as alternative or adjunctive treatments (6). In the treatment of RNDF, 5 of 8 patients received oral PSL, all of whom showed favourable responses (1–5). Two of these were started on treatment with low-dose of PSL (15 mg/day) in combination with dapsone (3, 4). Follow-up data after PSL therapy were documented in 4 patients, 2 of whom showed recurrence while taking low-dose PSL (15 and 5 mg/day, respectively) (2, 3), whereas the other 2 had no recurrence after discontinuation or during tapering to 2 mg/day (4, 5). In addition, 1 patient was treated with oral erythromycin alone, which alleviated the lesions (1). The remaining 2 patients, including the current case, were treated with potassium iodide, and in both instances the lesions were alleviated within 1 month (4). In the current case, the lesions partially recurred 10 and 18 months after starting treatment, while in the other patient, the lesions partially recurred during tapering (dose unknown) (4). Both patients improved rapidly upon increasing the potassium iodide dose. Therefore, potassium iodide alone may be effective in patients with RNDF. Considering that treatment may be prolonged with recurrence and that potassium iodide has fewer side effects than PSL, we suggest that potassium iodide should be prioritized in RNDF treatment. The dose should be adjusted according to the severity of clinical symptoms, with attention to side effects, including drug-induced thyroid disorders. Oral PSL should be considered if potassium iodide is ineffective.

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