A case of lupus miliaris disseminatus faciei after allogeneic hematopoietic stem cell transplantation

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To the Editor: A 43-year-old man was diagnosed with acute T-cell lymphoblastic leukemia. He underwent allogeneic stem cell transplantation with a 6/6 human leukocyte antigen (HLA)-match; the donor was his sister. Cyclosporine A was used for 5 months as a prophylactic for graft *vs.* host disease. Three months after discontinuation of cyclosporine A, he developed papules on his face. Upon physical examination, multiple small dome-shaped, reddish-brown papules were observed on the central area of the face that extended to both cheeks, lower eyelids, and chin [Figure 1A]. Extra-facial lesions were absent. Dermoscopy examination of the papules demonstrated dotted vessels and linear vessels arranged both horizontally and vertically, forming a polygonal pattern, and the structure of targetoid follicule plugs was not observed. The mode of dermoscopy for observation was polarized light, non-wetting [Figure 1E]. Diascopic examination revealed an apple-jelly appearance. Histopathological analysis demonstrated epithelioid cell granulomas, with central areas of caseation necrosis and Langhans giant cells surrounding a moderate lymphohistiocytic infiltrate [Figure 1C and 1D]. An acid-fast stain was performed on the pathology tablets, and no mycobacteria were found. Laboratory analyses, including full blood count, erythrocyte sedimentation rate, serum calcemia, serum angiotensin-I-converting enzyme level, blood chemistry, tuberculosis infection T cell spot test (T-SPOT), and a chest X-ray were normal or negative. He was diagnosed with lupus miliaris disseminatus faciei (LMDF). The patient and the donor both had rosacea and LMDF before. In the first 3 months, intra-muscular injection of 1 mL of the compound betamethasone was given once a month. The patient was also prescribed isotretinoin at a dose of 10 mg/day and topical tacrolimus twice a day and had a good clinical response. After 6 months of treatment, the lesions improved [Figure 1B].

The etiology of LMDF remains unknown. It has been considered a variant of a *Mycobacterium tuberculosis* infection. However, the evidence supporting this theory has not been found.[1] In addition, no evidence of a tuberculous infection was observed in our case. LMDF is widely considered as a granulomatous rosacea. However, LMDF is different from granulomatous rosacea in that it has involvement of extra-facial sites, no vascular symptoms, and a self-limiting course with scar formation.[2] It seems highly likely that an immune response to the pilosebaceous units is involved in the granuloma formation in LMDF. It has been suggested that an allergic granulomatous reaction to foreign bodies or a reaction to follicular contents, such as keratins and sebum has led to its formation.[3] In 2015, Nishimoto *et al*[4] reported that cell-mediated immunity to *Propionibacterium acnes* (*P. acnes*) may play a role in granuloma formation. Cell-mediated immunity occurs when a hair follicle is destroyed and subsequently *P. acnes* enters into the dermis.

In this patient, histopathological examination revealed dermal epithelioid cell granulomas, some with central areas of necrosis, and surrounding a moderate lymphohistiocytic infiltrate; however, the tuberculosis laboratory tests were negative. Moreover, in this patient, LMDF occurred after discontinuation of cyclosporine A. It has been reported that in some patients with LMDF, there might be some hereditary or acquired excessive cellular immune response to *P. acnes*, which is similar to sarcoidosis. Our patient developed LMDF after allogeneic hematopoietic stem cell transplantation, thereby suggesting that the immunological derangement after transplantation plays a role in the pathogenesis of LMDF.

Oral isotretinoin and topical tacrolimus have been discussed as effective treatments. Several other approaches, including intra-lesional or systemic corticosteroids, tetracyclines, metronidazole, erythromycin, dapsone, antituberculous antibiotics, chloroquine, and clofazimine were attempted with variable results.[5] In our case, the patient was treated 6 months with oral isotretinoin and topical tacrolimus, and achieved good results.

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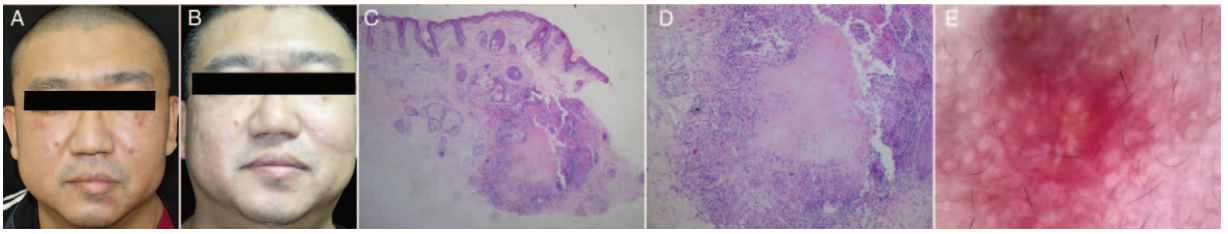

Figure 1: The clinical manifestation, pathological and dermoscopic findings of the patient. (A) Clinical findings at the time of the initial examination. Multiple small dome-shaped, reddish-brown papules were present on the central area of the face that extended to both cheek, lower eyelids, and chin. (B) After 6 months of treatment with oral isotretinoin and topical tacrolimus, the papules had mostly disappeared with scarring. (C) Dermal epithelioid cell granulomas, some with caseation necrosis, and surrounding a moderate lymphohistiocytic infiltrate with multinucleate giant cells, mostly of the Langhans-type (hematoxylin-eosin staining, original magnification ×40). (D) Histiocytes and Langerhans-type multinucleate giant cells are visible as encircling the caseation necrosis, with a peripheral rim of lymphocytes (hematoxylin-eosin staining, original magnification ×100). (E) Dermoscopic examination of papules showing dotted vessels and linear vessels arranged horizontally and vertically, forming a polygonal pattern (original magnification ×200).

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the article. The patient understands that his name and initials will not be published and due efforts will be made to conceal the identity of the patient, although anonymity cannot be guaranteed.

Conflicts of interest

None.

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