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Trichostasis Spinulosa as Manifestation of Cutaneous Graft versus Host Disease

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Dear Editor:

A 55-year-old male was diagnosed with CD20-positive acute B-lymphoblastic leukemia and was in complete remission after chemoimmunotherapy with cyclophosphamide, vincristine, doxorubicin and rituximab. He underwent a haploidentical haematopoietic stem cell transplant with a conditioning regime of intravenous (IV) fludarabine/thiotepa/melphalan/total lymphoid irradiation. The transplant was complicated by acute graft versus host disease (GVHD) manifesting with gastrointestinal tract involvement on Day 25 post-transplantation. A colon ulcer biopsy confirmed the diagnosis of GVHD histologically—the large bowel mucosa showed crypt distortion and loss with ulceration, with apoptotic bodies and "exploding crypts" seen in the base of crypts. On Day 20 post-transplant, cutaneous examination revealed minimally keratotic follicular papules distributed extensively over the extensors of arms and legs (Fig. 1A). These papules had central short protruding dark hairs. Clinical impression was that of trichostasis spinulosa. Histology revealed perifollicular fibrosis and dilation of hair follicles (Fig. 1B). There was hyperkeratosis and eccentric atrophy of hair follicle epithelium in the infundibular region, associated with focal vacuolar interface change and pigmentary incontinence. A few hair follicles contain at most two hair shafts on multiple levels of section examined. Histological hair-follicle damage was compatible with cutaneous GVHD. Clinical-pathological correlation led to a diagnosis of trichostasis spinulosa-like changes in GVHD. This patient had GVHD involving the gastrointestinal tract with GVHD changes in skin. Hair changes reported in acute GVHD include erythematous-to-hyperpigmented follicular papules, present on the shoulders, lateral thorax, and anterior thighs[1]. Histological findings include dyskeratosis of individual keratinocytes, pat-

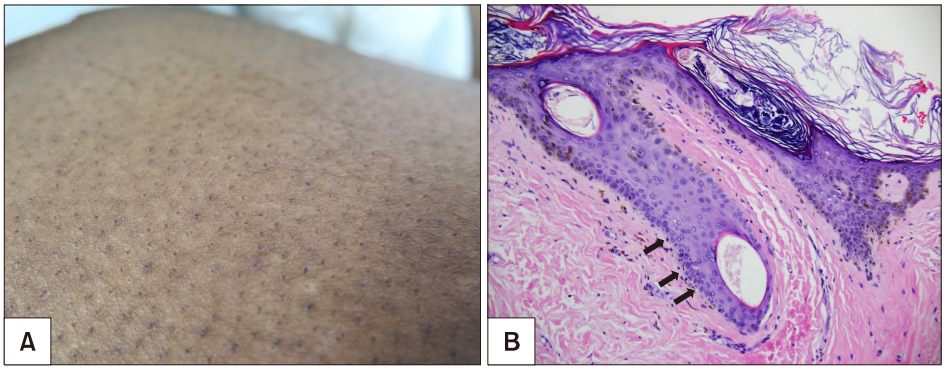

Fig. 1. (A) Minimally keratotic follicular papules with central short protruding dark hairs distributed extensively over the extensors of arms and legs. (B) Photomicrograph showing dilated hair follicles with eccentric atrophy of hair follicle epithelium, focal vacuolar interface change (indicated by arrows) and pigmentary incontinence (H&E, ×200).

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chy vacuolization of the basement membrane, dermal and perivascular lymphocytic infiltrate[2]. Hair changes are more prominent and diverse in chronic GVHD, including scarring or non-scarring alopecia, scaly scalp papules, thinning hair, coarse hair, and premature greying[3]. Follicular involvement in the form of follicular keratosis is also increasingly reported in both lichenoid[4] and early phases of sclerodermatous chronic GVHD. The 2014 NIH Consensus classifies keratosis pilaris in the category of rare, controversial, or nonspecific features that cannot be used to establish the diagnosis of chronic GVHD[3]. In trichostasis spinulosa, multiple tiny short hairs become embedded within hair follicles, with resultant dark, spiny papules on the face or trunk. The classical variant presents with non-itching, comedo-like lesions on the face in the elderly, while the pruritic variant presents with itching, follicular papules located on the limbs in young adults[5]. The later variant is often confused with keratosis pilaris. Histology of trichostasis spinulosa typically shows hyperkeratosis with follicular plugging, a widened hair follicle, perifollicular inflammation, and multiple vellus hairs enveloped in a keratinous sheath. Clinical features determine whether the clinical syndrome of GVHD is considered acute or chronic, not the temporal relationship to transplantation[3]. We propose that our patients had hair changes similar to trichostasis spinulosa, which is regarded as a new finding of cutaneous GVHD. The patient passed away 1 month after the diagnosis of GVHD. We highlight this case to add to the repertoire of dermatological manifestations in GVHD.

We received the patient's consent form about publishing all photographic materials.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

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