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Correspondence

Dermatitis herpetiformis arising within vitiligo in a patient with autoimmune polyendocrine syndrome type 3

The association between dermatitis herpetiformis (DH) and vitiligo is infrequent. It concerns 1.3% of the patients with DH[1] and 1% of the patients with vitiligo.[2] Case reports showed a peculiar affinity of DH lesions for vitiligo patches.[3–9] We report here an illustrative case of DH that displayed affinity phenomenon on vitiligo patches in a patient with autoimmune polyendocrine syndrome (APS) type 3.

A 47-year-old woman presented with a 2-week history of pruritic papulovesicular rash. Her past medical history included type 1 diabetes, hypothyroidism, vitiligo, asthma, hypertension, atopic eczema, and mood disorder. Her two children had celiac disease. The patient had a previous blood screening test with positive tissue transglutaminase IgA antibodies (19; normal <7). She denied any intestinal symptoms, and past gastroscopy and biopsies of the duodenum were normal. She had been followed in the past in our clinic for an itchy rash. Despite recurrent suspicions of DH, direct immunofluorescence (DIF) remained negative on several occasions 20 years ago. The rash had been treated since as atopic eczema with topical corticosteroids and moisturizers. Upon examination, the patient had symmetrical pruritic erythematous papules, vesicles, and excoriations on extensor surfaces of elbows, knees, and buttocks. She also displayed lesions on wrists, ankles, pubic area, and hairline (Fig. 1). The lesions were mainly localized within vitiligo. Vitiligo patches on the inner thighs and abdomen were mainly spared. DH disease was again highly suspected. A punch skin biopsy of a vesicle showed upper dermal inflammation with lymphocytes, macrophages and eosinophils, and subepidermal, cleavage. Two DIF from an erythematous area and a normal appearing perilesional area revealed granular IgA deposition on the basal

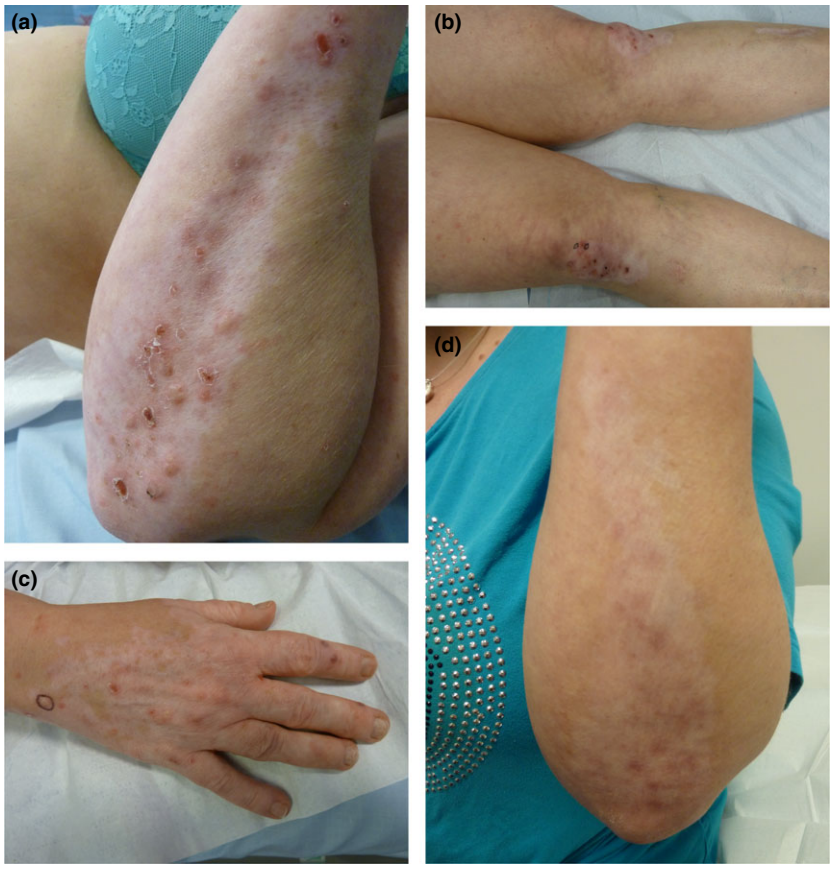

Figure 1 Vesicules of DH restricted to vitiligo of the elbows and forearm (a), knees (b), and dorsum of the hand (c). Complete regression after 3 months with dapsone 100 mg/day and gluten-free diet (d)

Table 1 Reported cases of dermatitis herpetiformis associated with vitiligo

Case	Author	Gender	Age (years) at the time of diagnosis of DH	Co-localization	First pathology	Other autoimmune conditions
1	Allende *et al.*, 1964[3]	M	35	No	Vitiligo	No
2	Ortonne *et al.*, 1976[2]	–	–	–	–	–
3	Olholm-Larsen *et al.*, 1980[4]	W	33	Yes	DH	No
4	Combs *et al.*, 1980[5]	M	25	Yes	Vitiligo	MGN
5	Hogan *et al.*, 1986[6]	W	46	Yes	Vitiligo	Antithyroid antibodies without hypothyroidism
6	Reunala *et al.*, 1997[1]	W	–	–	–	–
7	Reunala *et al.*, 1997[1]	M	–	–	–	–
8	Reunala *et al.*, 1997[1]	M	–	–	–	–
9	Reunala *et al.*, 1997[1]	M	–	–	DH	–
10	Amato *et al.*, 2000[7]	W	53	Yes	Vitiligo	Antithyroid antibodies without hypothyroidism
11	Karabudak *et al.*, 2007[8]	M	21	Yes	Vitiligo	No
12	Macbeth *et al.*, 2013[9]	W	37	Yes	Vitiligo	APS-2 (thyroiditis, adrenal insufficiency, DT1, CD)
13	Present case, 2018	W	47	Yes	Vitiligo	APS-3 (DT1, thyroiditis, vitiligo)

APS, autoimmune polyendocrine syndrome; CD, celiac disease; DT1, type 1 diabetes; M, man; MGN, membranous glomerulonephritis; NA, not available; W, woman.
[a]Cases extracted from large series. No specific clinical data given.

membrane. She started oral disulone 100 mg/day along with gluten-free diet. No gastroscopy was performed. At 3-month follow-up, the patient was clear of any lesion.

We report here a new case of association between DH and vitiligo. Our case is striking by the very strong autoimmune personal and familial context. The patient had APS type 3, which is defined by autoimmune thyroiditis with other organ-specific immunity, here type 1 diabetes and vitiligo, but without adrenal insufficiency.[10] A previous case was reported with APS type 2.[9] The localization and affinity of DH lesions within vitiligo is striking. We found 12 other cases of DH coexisting with vitiligo in the literature (Table 1).[1–9] Half were women, and the median age at diagnosis of DH was 36 years. In most of the cases (7/9), vitiligo preceded DH. DH was co-localized totally or at least partly on vitiligo in 7/8 patients. Only Allende *et al.* reported a case of DH sparing vitiligo patches.[3] Additional autoimmune features were found in 5/8 patients with either APS (2/8), circulating antithyroid antibodies (2/8), or in one case membranous glomerulonephritis during follow-up.[5] The mechanism of DH affinity for vitiligo patches is unknown. Hypotheses of the literature include: (i) Köebner phenomenon,[4] (ii) genetic mosaicism in case of segmental vitiligo,[9] (iii) fortuitous as DH lesions may be located on other areas without vitiligo.[8] Besides, DH antigens could be unmasked in vitiliginous areas under the action of UV exposure, trauma, or modified local immunity associated with melanocyte destruction.

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