

Systematized Keratotic Nevus with Finger Contracture

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A case of systematized keratotic nevus which is a peculiar form of epidermal nevus is reported. The present case is rare because of the dysfunction of incomplete flexion of fingers by systematized keratotic nevus. Furthermore, it involved mixed clinical and histological changes of epidermal nevus from fingers to elbow. An operation with full-thickness skin graft was undertaken. Key word: Mixed changes in epidermal nevus.

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Epidermal nevi are usually circumscribed hamartomatous lesions comprised predominantly of keratinocytes (1). There are some variations of this entity, both clinically or histologically (1–3). However, there are few published reports on systematized keratotic nevus. Patients with finger contracture due to systematized keratotic nevus are particularly rare. We describe a patient who had an unusual form of keratotic nevus.

CARE REPORT

A 28-year-old man was referred to us for evaluation of a papillomatous hyperkeratotic lesion on his left hand and forearm. According to his mother's statement, he had a rough brown lesion extending from the dorsum of the left fingers to the left forearm at birth. When he was 10 years old, papillomatous hyperkeratotic lesions gradually developed in parts of the rough brown lesion on his left hand, especially on the fingers. He had not sought treatment so far. He was the product of an uncomplicated pregnancy and normal spontaneous vaginal birth. There was no other anomaly and no known family history of a similar skin lesion.

Examination of the patient disclosed a brown lesion with

lichenoid appearance, extending from the dorsum of the left hand to the extensor surfaces of the left forearm. The brown pigmentation became paler from hand to elbow. Furthermore, there were papillomatous hyperkeratotic lesions on the dorsa of the thumb to middle finger and on the dorsum of the left hand (Fig. 1). He complained of the dysfunction of incomplete flexion of his left index and middle fingers.

A biopsy specimen from the papillomatous hyperkeratotic lesion of the thumb revealed pronounced hyperkeratosis, acanthosis with elongation of rete ridges and hypergranulosis (Fig. 2 a). There were many vacuolated cells from the granular to prickle cell layers. Cells in some parts of the granular layer contained enlarged keratohyalin gran-



Fig. 1. Papillomatous hyperkeratotic lesions and brown lichenoid lesions in the left hand.

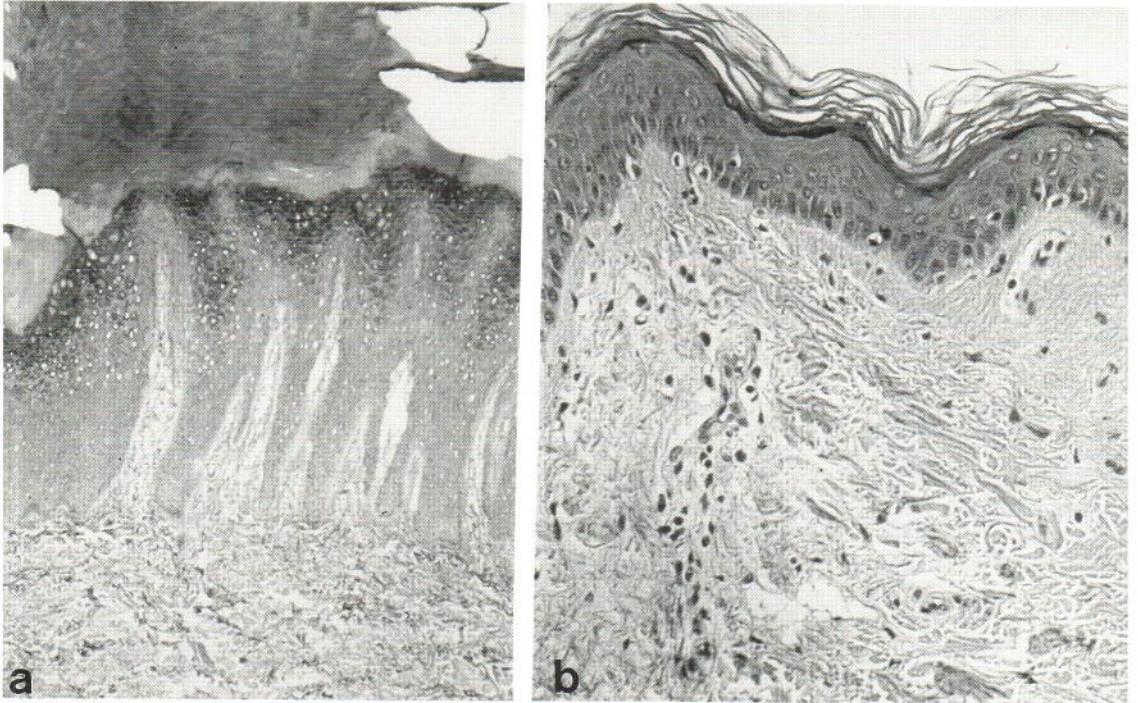


Fig. 2 a-b. (Left) Marked hyperkeratosis, acanthosis with elongation of rete ridges, hypergranulosis and granular degeneration are present in the papillomatous hyperkeratotic lesions (hematoxylin-eosin, original magnification $\times 40$). (Right) Slight hyperkeratosis and basal melanosis in the brown lichenoid lesion (hematoxylin-eosin, original magnification $\times 40$).

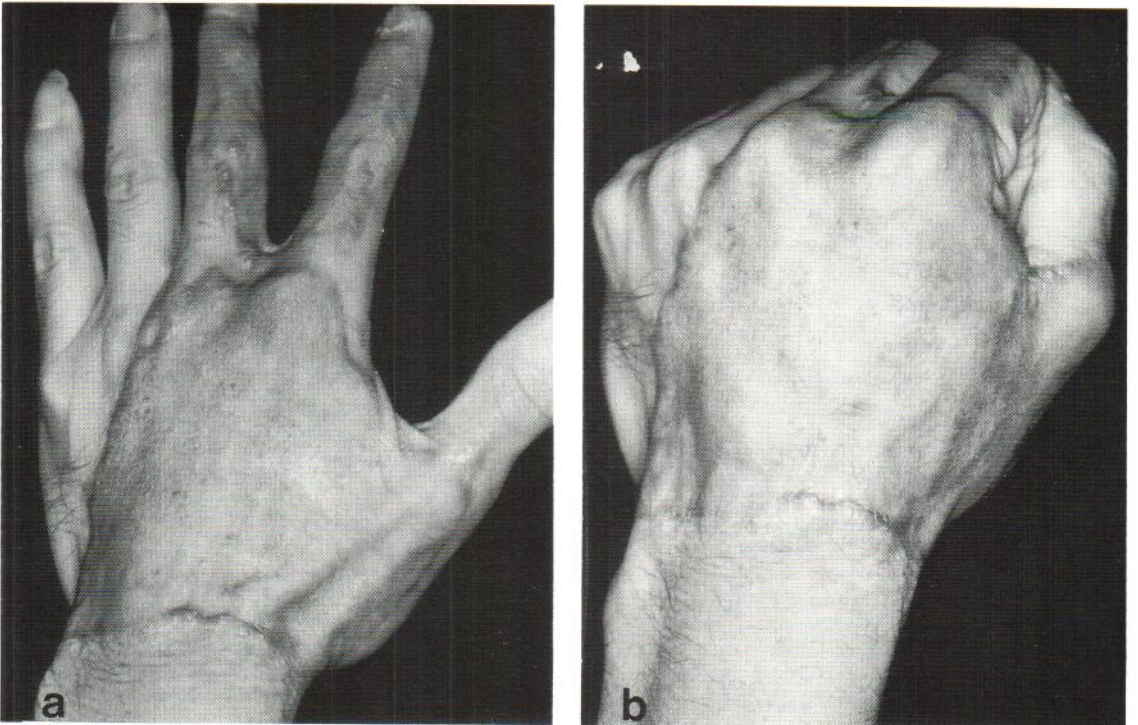


Fig 3 a-b. (Left) Six months after the operation of full-thickness free skin graft. (Right) Improved grasping.

ules in edematous cytoplasm suggestive of granular degeneration. By contrast, a biopsy specimen from the brown lichenoid on the left forearm showed slight hyperkeratosis and basal melanosis (fig. 2 b). Immunoenzymatic stainings for papilloma virus were negative in both biopsy specimens.

Based on the clinical and histological findings the lesion was diagnosed as keratotic nevus. The lesion on the left hand was excised distally from the left wrist and reconstructed with a full-thickness free skin graft (Fig. 3 a). His grasp is improved, 6 months after the operation (Fig. 3 b).

DISCUSSION

We diagnosed our case as a peculiar form of epidermal nevus; systematized keratotic nevus or nevus unius lateris. Though granular degeneration is frequently seen in systematized linear epidermal nevus (3) or keratotic nevus (4) histologically, the etiology remains unknown. The present case was rare because of finger contracture caused by the epidermal nevus. We have treated 60 patients with epidermal nevus over the past 10 years. None of them complained of such kind of dysfunction. Furthermore, our case was rare because of the mixed clinical and histological changes, which were a mixture of papillomatous hyperkeratotic lesions with granular degeneration and pigmented lichenoid lesions showing hyperkeratosis and basal melanosis.

Epidermal nevi are generally flat at first, becoming increasingly velvety over a period of years, then verrucous (1). They tend to gradually extend at the same time. In our case, however, only parts of the lesion of epidermal nevi proliferated to give a severe hyperkeratotic appearance. There might have been a special condition or irritation in this area. On the other hand, it is now known that epidermal proliferation and differentiation is, to a great extent, promoted by the dermis (5). The dermis of the affected fingers may influence both the contracture of the fingers and the hypertrophy of overlying epidermis.

REFERENCES

1. Atherton DJ, Rook A. Verrucous epidermal naevus. In: Rook A, Wilkinson DS, Ebling FJG, et al., eds. *Textbook of Dermatology*, 4th edn. Boston: Blackwell, 1986: 168-171.
2. Domonkos AN, Arnold HL, Odom RB. Epidermal nevi and tumors. In: *Diseases of the Skin*, 7th edn. Philadelphia: WB Saunders, 1982: 792-795.
3. Lever WF, Schaumber-Lever G. Linear epidermal nevus. In: *Histopathology of the Skin*, 6th edn. Philadelphia: JB Lippincott, 1983: 472-474.
4. Fukushiro R. Studies in hard nevi, particularly on a peculiar form of keratotic nevi. *Jpn J Dermatol* 1950; 60: 147-171.
5. Briggaman RA. Control of differentiation of epidermal structures. *Birth Defects* 1981; 17: 39-60.