A Case of Verrucous Hemangioma

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We report a case of a 6-month-old female who had had verrucous hemangioma since birth. The lesions were dark red, verrucous surfaced, hyperkeratotic papules on the right sole. A histopathological examination showed hyperkeratosis, papillomatosis, irregular acanthosis, lobular proliferation and dilatation of blood vessels in the deep dermis with a gap devoid of vessel proliferation in the mid dermis. (Ann Dermatol 10:(2) 143~146, 1998).

Key Word: Verrucous hemangioma

Verrucous hemangioma is a rare vascular malformation, a structural variant of capillary or cavernous hemangioma, in which reactive epidermal changes develop secondarily. In the literature, it is variously described. However, in 1967 Imperial and Helwig¹ introduced the new term verrucous hemangioma based on clinicopathological findings. Verrucous hemangioma usually involves the lower extremities and its onset is near the time of birth, or less frequently, in early infancy. The clinical appearance of verrucous hemangioma changes with time. Initially, it is indistinguishable from capillary hemangioma. With time, it increases in volume, the color becoming brown and the skin surface becoming verrucous¹.

However, we herein report a case of verrucous hemangioma with the initial presentation of a verrucous appearance from birth. To our knowledge, reports of a similar initial presentation are not mentioned in the literature.

CASE REPORT

A 6-month-old female baby was brought to our department because of dark red, verrucous surfaced papules measuring approximately 0.7 to 1 cm in diameter on the right sole (Fig. 1). The lesions

patient was not ambulatory. The family history and past medical history were not significant. A physical examination did not reveal any other vasculopathy or cutaneous lesions. The patient's general condition and developmental state were good. A complete blood count, urinalysis, and foot roentgenogram were all normal.

A 3 mm punch biopsy specimen of the largest

were present at birth. There was no history of

bleeding from trauma, scratching, or pressure. The

A 3 mm punch biopsy specimen of the largest papule revealed irregular acanthosis and papillomatosis of the epidermis with marked hyperkeratosis. The papillary dermis contained large vascular cavities surrounded by the rete ridges (Fig. 2). Although the biopsy specimen was not deep enough to provide histological information about the subcutaneous tissue, it showed lobular proliferation and dilatation of blood vessels in the deep dermis with a gap devoid of vessel proliferation in the mid dermis (Fig. 3).

We recommended total excision in order to prevent recurrence, scarring and gait disturbance. However, the treatment was refused because of the patient's age. At a follow up examination after 18 months, the size and extent of the lesions had not changed.

DISCUSSION

Verrucous hemangioma is a rare form of vascular malformation usually of the lower extremities that is usually congenital and only rarely occurs later in life.^{1,2}. It is typically solitary, but a case of multiple

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Table 1. Clinical features of verrucous hemangioma in the Korean literature.

| Case | Age(yr)/ Sex | Size(cm) | Onset | Site | Symptom | Presentation | Treatment & Outcome | Referen ce |
|------|-----------------|---------------------------|-------|-----------|------------------------|---|----------------------------|-----------------|
| 1 | 22/M | 3-4×6-7 | 6mon | Rt. leg | Itching, Tenderness | Erythematous macule, gradually becoming verrucous and dark blue | E&D and Cryotherapy | 6 |
| 2 | 13/M | coin to egg | birth | Lt. knee | None | Erythematous patch, gradually becomes a verrucous surfaced and necrotic crusted plaque | Cryotherapy | 7 |
| 3 | 22/M | walnut | birth | Rt. knee | None | Erythematous macule, gradually becomes a verrucous surfaced and dark red nodule | | 8 |
| 4 | 20/M | $1.5 \times 6 \times 0.7$ | birth | Lt. foot | None | Erythematous macule, gradually becomes a dark red linear verrucous lesion | | 8 |
| 5 | 10/F | 2×2 | birth | Rt. leg | None | Reddish hemangiomatous lesion, becomes a verrucous crusty lesion | Lost to follow | 9 |
| 6 | 20/F | 1.8×2.2 | birth | Rt. leg | None | Reddish hemangiomatous lesion, becomes a verrucous crusty lesion | Excision and skin graft | 9 |
| 7 | 18/M | 2×6 | birth | Lt. leg | None | Erythematous patch, gradually becomes a hyperkeratotic and dark brown nodule. Several episodes of bleeding after trauma | Excision and skin graft | 10 |
| 8 | 23/M | 2×2 | birth | Lt. thigh | None | | Excision and skin graft | 10 |
| 9 | 15/M | $10\times6\times0.8$ | birth | Rt. thigh | None | Erythematous macule, gradually becomes a dark red, verrucous surfaced, hemangiomatous tumor | Excision | 11 |
| 10 | 6 mon/F | 0.7 to 1 in diameter | birth | Rt. sole | None | Dark red, verrucous surfaced plaque | Refused treatment | present case |

E&D: electrodesiccation

eruptive verrucous hemangiomas³ has been reported. Although digital verrucous fibroangioma⁴, which is a variant of verrucous hemangioma, usually appears as dome-shaped nodules at birth, most cases initially appear as an asymtomatic, flat, erythematous macule which slowly enlarges with age and becomes a wart-like, dark blue papule or nodule later in life, usually following repeated trauma and infection. Loria et al.5 suggested that the lesions pass through a basic hemangiomatous phase, followed by a progressive hyperkeratotic phase. The verrucous and hyperkeratotic features of the lesions are regarded as a reaction to injury, or possibly to an altered physiological state. However, our case initially presented with wart-like dark red papules without any trauma or infecton. This finding is different from the view of pathogenesis previously proposed. Clinical features of 9 cases from the Korean literature are presented in Table 16-11. The patient age at presentation, except in our case, ranged from 10 to 23 years(mean 18). All the reported cases began either as erythematous macules or patches, gradually becoming verrucous following repeated trauma or infection. In the clinical features of 10 cases reported by JKC Chan et al. 12, the patient age at presentation ranged from 6 to 44 years(mean 22. 7). Patient age at presentation ranged from 12 to 43 years in the clinicopathological study of 21 cases reported by Imperial et al.¹. We found no explanation in the literature as to why most reported lesions are macules or patches, while our case initially appeared as a verrucous plaque. Further evaluation is needed.

The histopathological findings of verrucous he-

Fig. 1. Dark red, verrucous surfaced papules on the right sole.

Fig. 3. Lobular proliferation and dilatation of blood vessels in the deep dermis (H & E stain, $\times 100$).

mangioma are a combination of congested capillaries and cavern-like vascular spaces in the superficial deep dermis and subcutaneous tissue with a hyperplastic epidermis showing variable degrees of acanthosis, papillomatosis, and hyperkeratosis. Clinical similarities to verrucous hemangioma are observed in angiokeratoma circumscriptum naeviforme(Acn), blue rubber bleb nevus and verrucous epidermal nevus. Confusion has occurred in the separation of Acn from verrucous hemangioma. However, the most important difference is apparently blood vessel proliferation in the dermis and subcutaneous tissue, whereas in Acn the papillary dermis shows only numerous, dilated, thin-walled vessels¹³.

Verrucous hemangioma does not regress spontaneously and enlarges with age, so it should be excised at a young age to prevent scarring and gait disturbance.

Fig. 2. Epidermis shows irregular acanthosis and papillomatosis, and papillary dermis contains large vascular cavities surrounded by rete ridges. (H & E stain × 40).

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