

Acquired Multiple Cavernous Hemangiomas of the Hand

Sir,

Cavernous hemangiomas are dermal or subcutaneous tumor-like aggregates of dilated vessels, which are lined by mature endothelial cells and further surrounded by thickened fibrous components (1). They usually appear in infancy as a solitary nodule and persist indefinitely. It is very rare to see multiple lesions in the skin. We report here a case of acquired multiple cavernous hemangiomas of the hand, uncomplicated by any association with a systemic disorder.

CASE REPORT

A 37-year-old woman consulted our clinic in June, 1993, for twelve bluish nodules on her left hand, which had a history of approximately 18 months. The patients's birth and health until that time were apparently normal. The first tumor appeared on the dorsum of the left index finger without history of trauma, and thereafter tumors gradually increased in size and number. There was no spontaneous pain or tenderness. Physical examination revealed twelve subcutaneous nodules on the left hand and fingers (Fig. 1A) with the largest nodule being 2 cm in diameter (Fig. 1B). Each tumor was dome-shaped, poorly defined and

bluish in color. The larger tumors were soft, while the smaller ones were hard. There were no other tumors or nevi found in any other sites. A blood test did not show any abnormal findings, and roentgenograms of the left hand and extremities did not show any bone involvement. After incisional biopsy, all the tumors were excised. With hematoxylin and eosin staining, the twelve tumors were found to be localized mainly in subcutaneous spaces without the involvement of muscle or tendon. The soft tumors consisted of a large mass of dilated vessels surrounded by thickened fibrous components (Fig. 2A), which is compatible with the history of cavernous hemangioma. The hard tumors consisted of large concentric fibrous nodules surrounded by capillaries (Fig. 2B). Thickened fibrous components surrounding the vessels and concentric fibrous nodules were stained blue with azan-Mallory staining but did not stain for desmin, thereby suggesting that they were collagen fibers. Endothelial cells of each lumen were positive for vimentin and Factor VIII-related antigen.

DISCUSSION

Solitary cavernous hemangioma is a common disease in the dermatological field (1); however, a case of acquired cavernous hemangioma with more than two lesions is very rare. Although

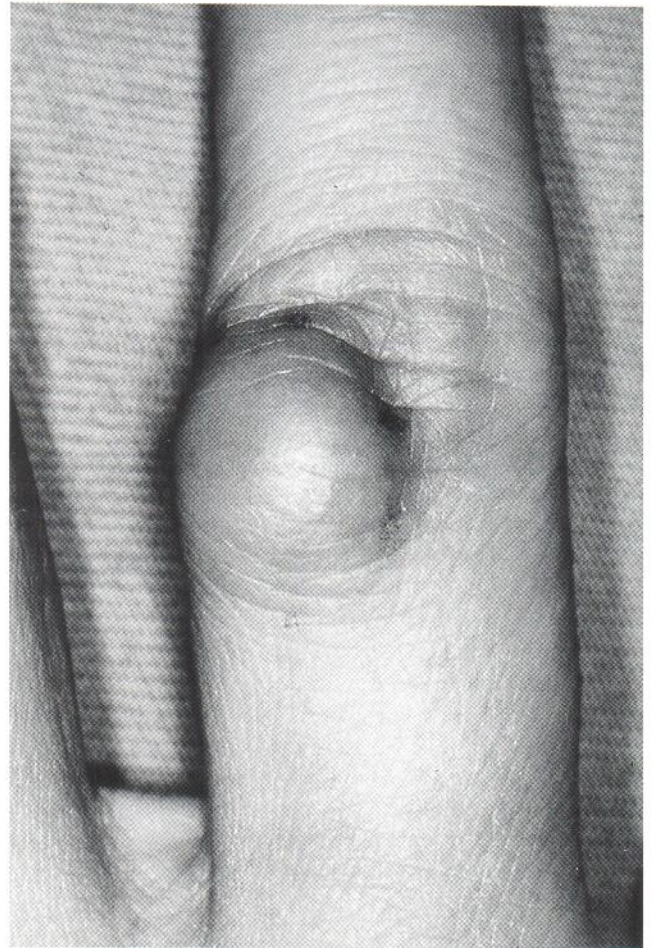
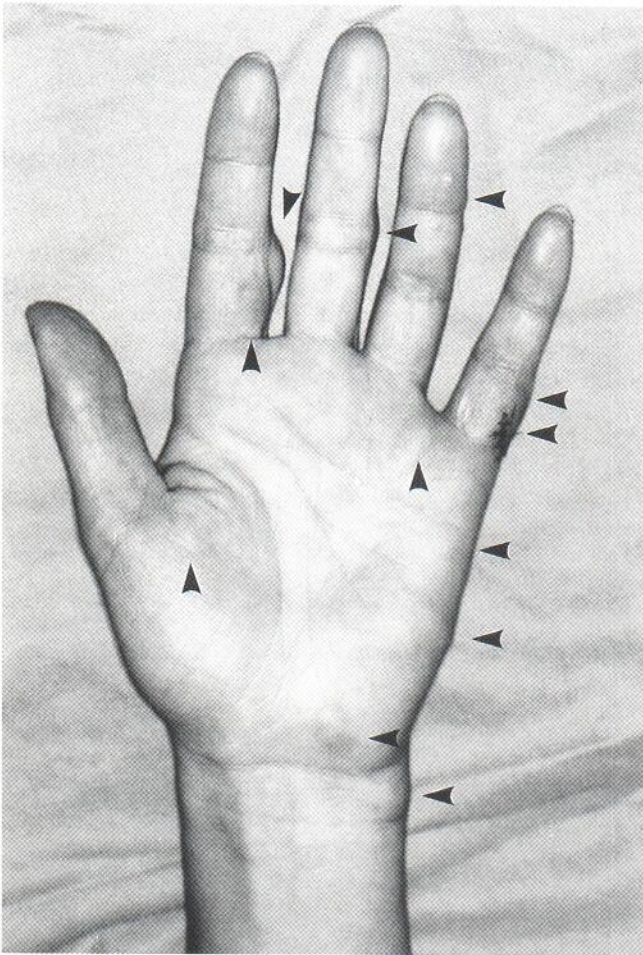


Fig. 1. A. Clinical manifestations of the subcutaneous tumors of the left hand. Arrows indicate some of the tumors. B. A subcutaneous tumor on the left index finger.

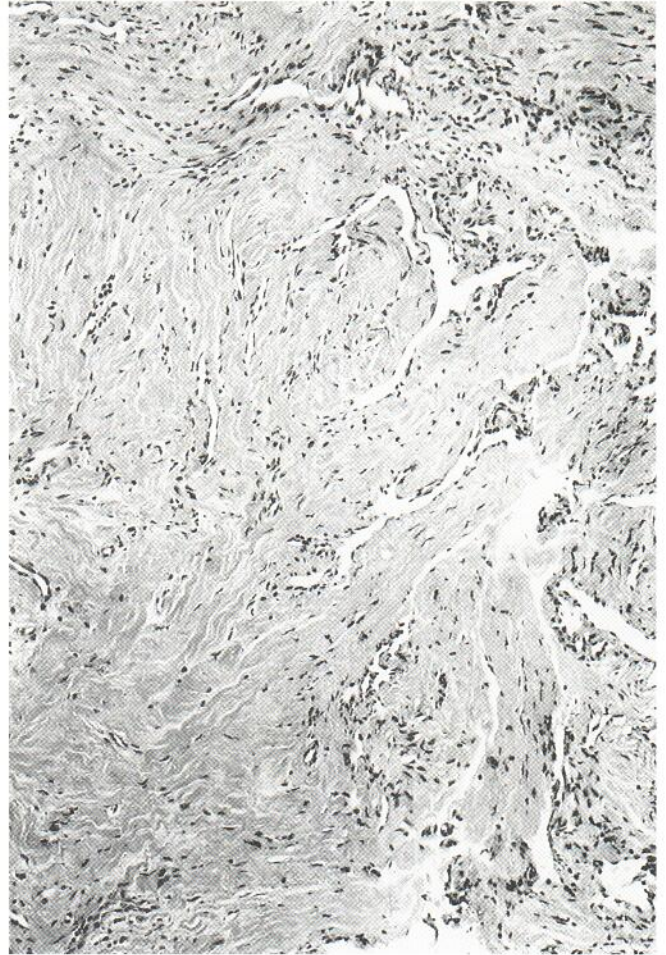
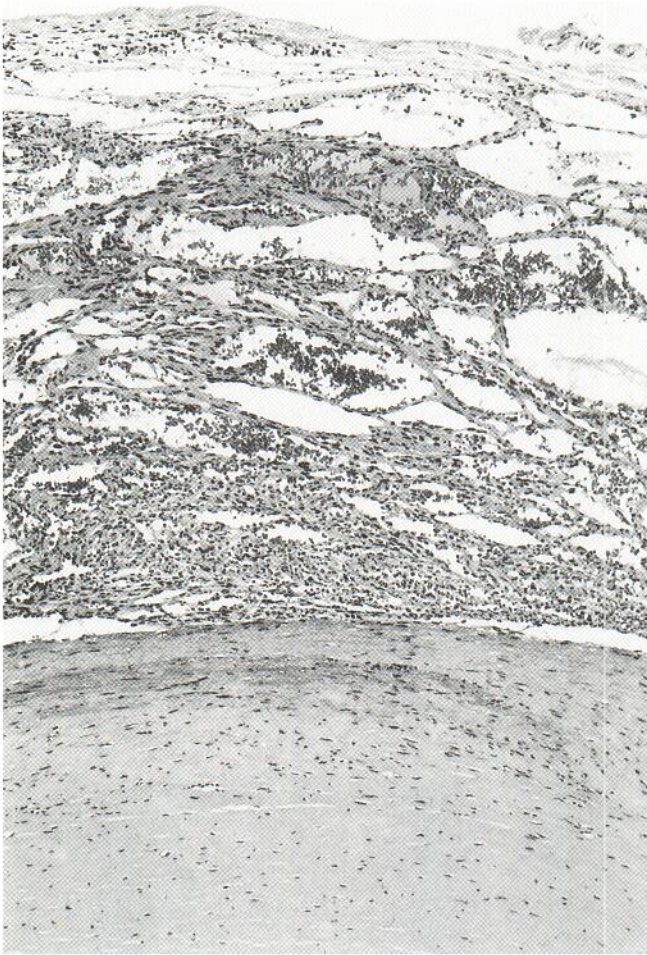


Fig. 2. A,B. Hematoxylin and eosin staining of the tumors (A: $\times 110$, B: $\times 83$).

multiple hemangiomas are observed in "Maffucci's syndrome" and "blue-rubber-bleb nevus syndrome" (1), our patient had never experienced any systemic symptoms. Furthermore, since the patient's hemangiomas first appeared at middle age with a relatively short onset at a localized site, the hemangiomas can be assumed not to be related to phacomatosis. Within the orthopedic field, hemangioma of the hand (2-4) is considered relatively common, with ganglions, giant cell tumors of the tendon sheath and mucous cysts being more frequent (2). Hemangioma of the hand located in a subcutaneous area is rarely congenital and more frequently occurs during adolescence (3), with surgical treatment being required when there is progressive enlargement with throbbing pain (2-4). Palmieri has reported that among 160 patients examined for hemangioma of the hand, 57% of the cases were capillary hemangiomas and 23% were cavernous hemangiomas (2), while only 10 patients (6%) had two separate lesions arising from different origins within the same hand (2). Examination of the English and Japanese literature has indicated that there may not yet have been any report published describing a case where acquired cavernous hemangiomas involved more

than two lesions. Although the precise cause of the lesion is not clear at present, a rich vascular network and frequent physical movement of the hand may be the basis for the occurrence of hemangiomas.

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Accepted December 15, 1994.

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*Reprint request