Sinusoidal Hemangioma In An Adult Male

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Citation

Abstract
Sinusoidal hemangioma is a rare variant of acquired cavernous hemangioma predominantly occurring in female. We are reporting such a case occurring in a 29-year-old man and briefly reviewing the literature.

Introduction
Calonje and Fletcher first reported sinusoidal hemangioma in 1991 (1). They described this acquired benign vascular lesion as a rare subset of cavernous hemangioma occurring mostly in adult females. The lesions were solitary, deep dermal or subcutaneous lobular vascular nodules histologically composed of dilated intercommunicating vascular channels lined by single layer of endothelial cells with occasional pseudopapillae formation. The purpose of this paper is to report such a case occurring in an adult male and to update the literature on this topic.

Case Report
A 29-year-old man noticed a slow-growing painless nodule over his left distal forearm for about one year. Clinical examination showed a non-tender, firm, slightly raised 1.5 cm subcutaneous mass withpurplish discoloration of the overlying skin. There was no other systemic disease. The patient underwent an excisional biopsy of the lesion. Gross examination showed a subcutaneous, well demarcated, lobulated mass measuring 1.0 X2.8 cm. Cut surface was pink tan and firm. Microscopically (Figs. 1 and 2), the tumor showed well-circumscribed lobules composed of vascular channels lined by flat and cuboidal endothelial cells. There were back-to-back dilated and congested vascular spaces with a prominent sinusoidal pattern. There were occasional minute thrombi noted in the blood vessels. Papillary endothelial hyperplasia was not noted. Several lobules showed central fibrosis. There was no significant nuclear atypia or mitoses of the endothelial cells.
Discussion
In the original article in 1991, Calonje and Fletcher described a distinctive variant of cavernous hemangioma and proposed that the entity be called sinusoidal hemangioma (1). A total number of 12 adult patients, eight female and four male, aged 20 to 77 years presented with solitary
subcutaneous or deep dermal lesions. The lesions were painless and slow growing over a period ranging from 3 weeks to 7 years before they were diagnosed. The lesions were located on the extremities (5 patients), trunk (5 patients), and breasts (2 patients). The lesions were treated by local excision. There were no recurrence or metastasis. Grossly the tumor varied in size from 1 to 3.5 cm. They were well circumscribed, spongy, and hemorrhagic. Microscopically, the deep dermis and the subcutaneous tissue showed a multilobular vascular tumor with a sieve-like appearance. The tumor was composed of dilated thin-walled vascular channels with area of pseudopapillary pattern. Single layer of endothelium, some showing focal pleomorphism and hyperchromatic nuclei were seen in a few cases from the breast raising the possibility of malignancy.

In 1998, Enjolras et al (2) described four cases, 2 males and 2 females with hemangiomas showing lobular spongiotic tumors composed of dilated, blood-filled, thin-walled vascular channels. Histology of the lesions was similar to that of sinusoidal hemangioma described by Calonje and Fletcher. However, clinically, the cases were very different. All the lesions were infantile or congenital in onset and had a highly aggressive and protracted clinical course. In 1999, Nakamura and Miyachi (3) reported an additional case of sinusoidal hemangioma occurring in a 43-year-old woman who presented with an enlarging lesion on her back over 20 years. A biopsy revealed the typical histology of sinusoidal hemangioma with areas of dystrophic calcification.

The present case that we have reported appears to be a typical sinusoidal hemangioma clinically and histologically as described by the original authors (1). The very few reports of such a lesion in the literature reflect either rarity of such lesions or unfamiliarity of this subset among the pathologists.

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References