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CASE REPORT

A Case of Sinusoidal Hemangioma with Lipoma

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Sinusoidal hemangioma is a distinctive subset of a group of lesions known collectively as cavernous hemangiomas. Clinically, it develops in adults, predominantly females, and presents as a solitary, painless, bluish, deep dermal or subcutaneous nodule. Lipoma is the most common benign soft tissue tumor. Lipoma is distinguished from sinusoidal hemangioma on both clinical and histological grounds. Several studies have suggested that adipocytes originate from perivascular cells during adipogenesis. Angiogenic cytokines released by adipocytes play a role in the vasoproliferative response. The rearrangement or loss of chromosome 13 can also be associated with hemangioma. However, no previous cases of sinusoidal hemangioma have been associated with benign tumors like lipoma. Here, we describe an unusual case of sinusoidal hemangioma that occurred together with a lipoma on the right upper arm of a 43-year-old male. **(Ann Dermatol 23(S2) S250～S253, 2011)**

-Keywords-
Angiogenic cytokine, Chromosome 13, Lipoma, Sinusoidal hemangioma

INTRODUCTION

Sinusoidal hemangioma is a distinctive subset of a group of cavernous hemangioma first described by Calonje and Fletcher[1] in 1991. Clinically, it develops in middle-aged adults, predominantly in females, and presents as a solitary, painless, bluish subcutaneous nodule. Although anatomic distribution is wide, the tumor presents most frequently on the extremities, trunk, and often on the breast and scalp[1]. Histologically, it is characterized by a well-circumscribed lobulated architecture composed of dilated interconnecting, thin-walled vascular channels that frequently show a pseudopapillary pattern and a back-to-back arrangement with scanty intervening stroma[1-3].

Lipoma is a common benign soft tissue tumor, that usually develops between the ages of 40 and 70 years. Histologically, lipomas contain mature adipocytes as a principle component. They tend to be surrounded by a thin capsule of connective tissue and are composed often entirely of normal fat cells indistinguishable from fat cells in the subcutaneous tissue[4].

Sinusoidal hemangioma is rarely reported. Here, we report on an interesting case of sinusoidal hemangioma with a lipoma on the right arm that occurred in a 43-year old male patient. To the best of our knowledge, this is the first report of a case with the combined tumors.

CASE REPORT

A 43-year-old male visited our clinic complaining of an asymptomatic solitary subcutaneous nodule on his right upper arm. He presented with a two-month history and claimed that he had noticed it by accident. On physical examination there were no symptoms except for a skin lesion. Skin examination revealed a solitary, relatively well-defined, pea-sized bluish subcutaneous nodule on the right upper arm (Fig. 1). There was tenderness on palpation, and no bleeding or ulceration was observed. All laboratory examinations, including a complete blood count, blood coagulation test, routine chemistry, and venereal disease research laboratory test were either in the normal range or negative. Histopathologically, there was a

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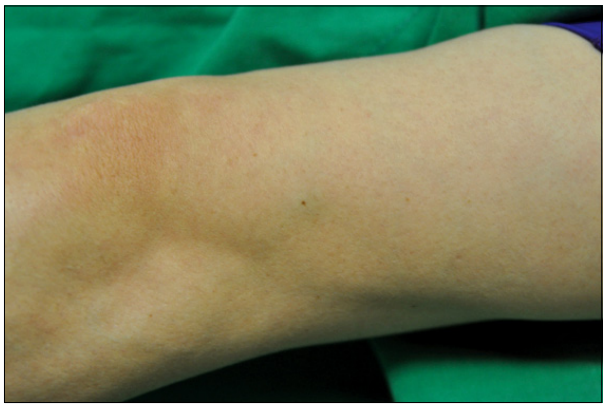

Fig. 1. A solitary, relatively well-defined, pea-sized, bluish subcutaneous nodule on the right upper arm.

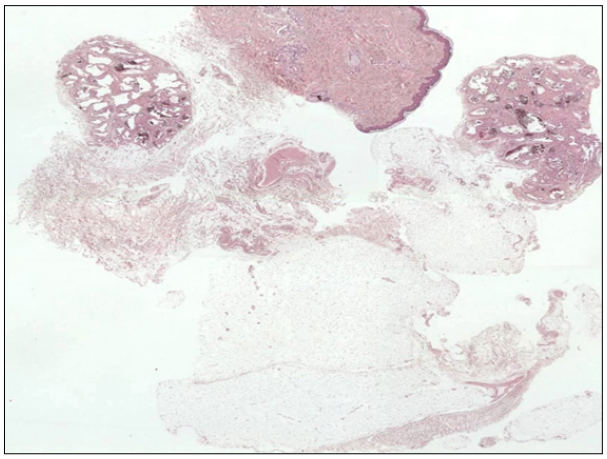

Fig. 2. On the upper left side of the picture, is a well-circumscribed, lobulated mass in the subcutaneous tissue. The mass is not encapsulated, and is composed of dilated and interconnecting vascular channels forming a sinusoidal structure. In the lower right region of the mass, is a well-circumscribed collection of benign mature adipocytes surrounded by a fibrous capsule (H&E, ×20).

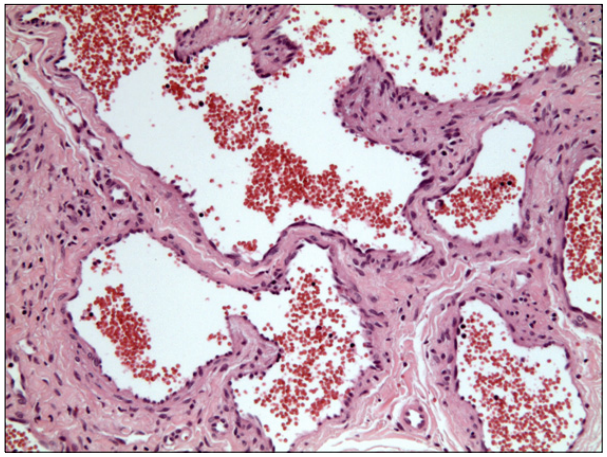

Fig. 3. Dilated vessels, with pseudopapillae and many red blood cells in the lumen, show a back-to-back arrangement (H&E, ×200).

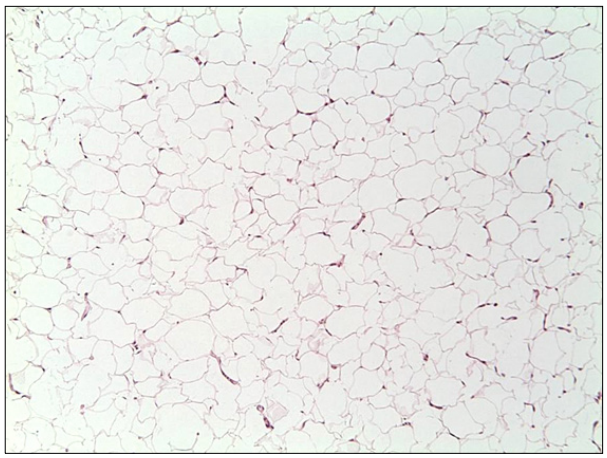

Fig. 4. In the lower right region of the hemangioma is, a benign tumor consisting of mature signet-ring fat cells (H&E, ×100).

well-circumscribed, lobulated subcutaneous mass. The mass was not encapsulated, and was composed of dilated and interconnecting, thin-walled vascular channels forming a sinusoidal structure. The channels were filled with many red blood cells. On the right side of the mass, was a well-circumscribed collection of benign mature adipocytes (Fig. 2). The dilated vessels showed a pseudopapillary pattern and a back-to-back arrangement with scanty intervening stroma. The nuclei of the endothelial cells showed focal hyperchromasia and mild pleomorphism, but no mitotic figures (Fig. 3). In addition, the mature adipocytes were surrounded by a capsule of thin connective tissue. However, features of inflammation and malignancy were not seen (Fig. 4). Based on the clinical and histological findings, the patient was diagnosed with sinusoidal hemangioma associated with lipoma, after which a simple excision was performed. Follow-up for 1 month after excision revealed no tendency to either local recurrence or metastasis.

DISCUSSION

Sinus hemangioma is a relatively rare variant of cavernous hemangioma. A typical feature during histopathology is the presence of gaping, markedly dilated and congested, thin-walled, back-to-back vascular spaces in a sieve-like or sinusoidal arrangement. Pseudopapillary structures due to cross-sectioning of these spaces focally resemble intravascular papillary endothelial hyperplasia. The blood vessels are lined by bland, flat endothelial cells, which can be focally prominent and mildly pleomorphic. Thrombosis,

hyalinization, dystrophic calcification, and even areas of infarction can be seen in older lesions[1-3]. Since they are composed of dilated vessels in subcutaneous tissue, cavernous hemangiomas are considered in the differential diagnosis. Cavernous hemangioma differs principally in its tendency to appear in childhood, be of a larger size, and present on the upper body. Histologically, in contrast to sinusoidal hemangioma, a cavernous lesion has a non-lobular, poorly demarcated structure and no pseudo-papillary appearance[3]. Although the anatomical distribution of sinusoidal hemangioma is wide, tumors often present in the breast. In our case, cytological atypia and hyperchromasia were seen histologically in the endothelial and perivascular cells. Therefore, the need to differentiate this type of tumor from well-differentiated angiosarcoma and avoid unnecessary radical treatment was emphasized[1,3]. Angiosarcoma is an interstitial lesion and not a subcutaneous lesion. It typically shows diffuse infiltration, atypical mitosis and multilayered endothelial cells[3]. Another tumor that shows papillary projection is the intravascular papillary endothelial hyperplasia. This tumor contains eosinophilic interstitial stroma, which are not observed in sinusoidal hemangioma.

Another subcutaneous hemangioma, like subcutaneous pyogenic granuloma (lobular capillary hemangioma), spindle cell hemangioma and cellular angiolipoma are considered to be part of the differential diagnosis. The characteristic histopathologic features of subcutaneous pyogenic granuloma are distinctive lobules of dilated and congested capillaries. The angiomatous tissue is surrounded by myxoid stroma containing scattered spindle- and stellate-shaped connective tissue cells and occasional mast cells[5]. The lesion of spindle cell hemangioma is characterized by cavernous or slit-like vascular proliferations lined with a thin layer of endothelium, alternating with cellular areas consisting predominantly of spindle cells[6]. The histopathological findings of cellular angiolipoma show well-circumscribed tumors composed of vascular endothelial and spindle cells forming vascular spaces intermingled with scattered adipocytes[4,7]. Thus, it is possible to distinguish between these subcutaneous hemangiomas and sinusoidal hemangiomas.

Sinusoidal hemangioma combined with another benign tumor has never been reported. However Calonje and Fletcher[1] reported a case of sinusoidal hemangioma with metaplastic ossification while Nakamura and Miyachi[2] reported one that showed calcification and organized thrombi.

Lipoma is the most common benign mesenchymal neoplasm composed of mature adipocytes, and usually presents between the ages of 40 and 70 years. Most lipomas occur in subcutaneous tissue as superficial lipomas, and rarely in interosseous tissue as deep lipomas. Subcutaneous lipomas are frequently located on the trunk, and occasionally occur on the neck, forearm, and axilla. Histologically, they tend to be surrounded by a capsule of thin connective tissue and are composed often entirely of normal fat cells that are indistinguishable from fat cells in the subcutaneous tissue[4].

Two-months earlier, our patient had accidentally noticed a subcutaneous nodule on the right upper arm. Clinically it was suspected to be a lipoma, but was diagnosed histologically as a sinusoidal hemangioma combined with lipoma.

The pathophysiology of a sinusoidal hemangioma combined with lipoma has not been reported. Several recent electron microscopic studies suggest that adipocytes are closely related to capillaries and originate from perivascular cells, like prenatal adipogenesis[8]. From these studies, we believe that adipocytes from the perivascular cells, which make the sinusoidal hemangioma, also formed the lipoma. Also, as mentioned earlier, in the case of sinusoidal hemangioma with ossification, metaplastic lipoma can not be ruled out. Alternatively, the lipoma may have occurred accidentally from the abnormal condensation of adipocytes combined with the sinusoidal hemangioma.

Vascular endothelial growth factor (VEGF) and basic fibroblast growth factor (bFGF) have been demonstrated as regulators of angiogenesis[9]. These factors are found in higher concentrations and play a role in proliferative hemangiomas. Since adipose tissue serves as an important conduit for the vasculature, it is conceivable that the angiogenic properties of this tissue may modulate the growth of the vasculature in a paracrine manner. The angiogenic cytokines (FGF-2, VEGF) released by adipocytes and endogenous cytokines such as FGF-2 play a role in the vasoproliferative response[10].

The lipoma observed in our patient was able to stimulate angiogenesis. Previous cytogenetic studies showed that a substantial subset of adipose tissue tumors display a rearrangement or loss of chromosome 13[11]. Tharapel et al.[12] suggested that the hemangioma is a common features in distal trisomy 13. Maybe the rearrangement of chromosome 13 affected our patient. However, interconnections between the sinusoidal hemangioma and lipoma could not be specifically identified, and they may be a coincidence or another type of hemangioma. Further studies will be required to investigate possible interconnections.

REFERENCES

1. Calonje E, Fletcher CD. Sinusoidal hemangioma. A distinctive benign vascular neoplasm within the group of cavernous hemangiomas. Am J Surg Pathol 1991;15:1130-1135.
2. Nakamura M, Miyachi Y. Calcifying sinusoidal haemangioma on the back. Br J Dermatol 1999;141:377-378.
3. Kim EH, Kim YC, Lee SW. A case of polypoid sinusoidal hemangioma. Korean J Dermatol 2006;44:1016-1017.
4. Ragsdale BD. Tumors with fatty, muscular, osseous, and/or cartilaginous differentiation. In: Elder DE, Elenitsas R, Johnson BL Jr, Murphy GF, Xu X, editors. Lever's histopathology of the skin. 10th ed. Philadelphia: Lippincott Williams & Wilkins, 2009:1057-1106.
5. Lee EJ, Oh EJ, Jeong TJ, Shin MK, Kim NI. A case of subcutaneous pyogenic granuloma, and a review of the literature. Korean J Dermatol 2010;48:776-779.
6. Coras B, Hohenleutner U, Landthaler M, Hohenleutner S. Spindle cell hemangioma. Dermatol Surg 2003;29:875-878.
7. Kim SJ, Park HJ, Lee JY, Cho BK. A case of cellular angiolipoma. Korean J Dermatol 2008;46:1289-1291.
8. Dotz W, Prioleau PG. Nevus lipomatosus cutaneus superficialis. A light and electron microscopic study. Arch Dermatol 1984;120:376-379.
9. Chang J, Most D, Bresnick S, Mehrara B, Steinbrech DS, Reinisch J, et al. Proliferative hemangiomas: analysis of cytokine gene expression and angiogenesis. Plast Reconstr Surg 1999;103:1-9.
10. Lucarelli E, Sangiorgi L, Benassi S, Donati D, Gobbi GA, Picci P, et al. Angiogenesis in lipoma: an experimental study in the chick embryo chorioallantoic membrane. Int J Mol Med 1999;4:593-596.
11. Dahlén A, Debiec-Rychter M, Pedeutour F, Domanski HA, Höglund M, Bauer HC, et al. Clustering of deletions on chromosome 13 in benign and low-malignant lipomatous tumors. Int J Cancer 2003;103:616-623.
12. Tharapel SA, Lewandowski RC, Tharapel AT, Wilroy RS Jr. Phenotype-karyotype correlation in patients trisomic for various segments of chromosome 13. J Med Genet 1986;23:310-315.