Cutaneous Angiomyolipoma

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Angiomyolipoma is a benign tumor, almost exclusively of the kidney, that is often associated with tuberous sclerosis. Cutaneous location is extremely rare. We report a case of cutaneous angiomyolipoma which occurred on the retroauricular area. A 57-year-old man presented with an asymptomatic mass on the retroauricular area. Although the clinical similarity to an epidermal or pilar cyst was striking, the histopathologic picture was distinctive. The tumor was a well-circumscribed subcutaneous mass composed of a mixture of smooth muscle, vascular spaces, and mature fat covered by a thin fibrous membrane.
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INTRODUCTION

Angiomyolipoma is a rare, benign mesenchymal tumor that consists of a proliferation of blood vessels, smooth muscle bundles, and fat tissue. The tumor is frequently associated with tuberous sclerosis, and found in the renal parenchyme. Several cases of cutaneous angiomyolipoma have been reported since Argenyi et al.[1] provided the original case. In the Korean dermatologic literature, two cases of cutaneous angiomyolipoma have been reported[2,3]. We report an additional case of angiomyolipoma which developed on the retroauricular area.

CASE REPORT

A 57-year-old man presented to our outpatient clinic with an asymptomatic mass on the left retroauricular area which had been present for 4 years. Dermatologic examination revealed a 2.0 × 1.5 cm sized, smooth-surfaced, cystic mass on his left retroauricular area (Fig. 1). Clinically, the lesion appeared to be an epidermal cyst, and was completely excised. The hematoxylin and eosin-stained sections revealed a well-circumscribed subcutaneous mass that was surrounded by a thin layer of loose, fibrous connective tissue (Fig. 2A). Three components were found together in variable quantities within the lesion: mature adipose tissue, blood vessels, and smooth muscle (Fig. 2B). The fat cells were uniform in appearance with solitary cytoplasmic vacuoles of varying sizes and a small peripheral nucleus. Most vascular spaces were surrounded by a muscular coat, but occasional foci of thin-walled venules and capillary vessels were observed. The smooth muscle was arranged in interlacing fascicles intermixed with fat. In Masson's trichrome stained sections, muscular bundles stained red (Fig. 3). HMB-45 immunoreactivity was absent. Based on these features, the tumor was diagnosed as a cutaneous angiomyolipoma.

DISCUSSION

Angiomyolipoma is a benign tumor, mainly of the kidney, which is frequently associated with tuberous sclerosis. It has also occasionally been described in other locations, such as the vagina, retroperitoneum, liver, lymph nodes, nasal cavity and oral cavity[4]. Cutaneous location is extremely rare. In 1986, Argenyi et al.[1,5] provided the original report of a

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Fig. 1. A 2.0 × 1.5 cm sized, asymptomatic cystic mass on the left retroauricular area.

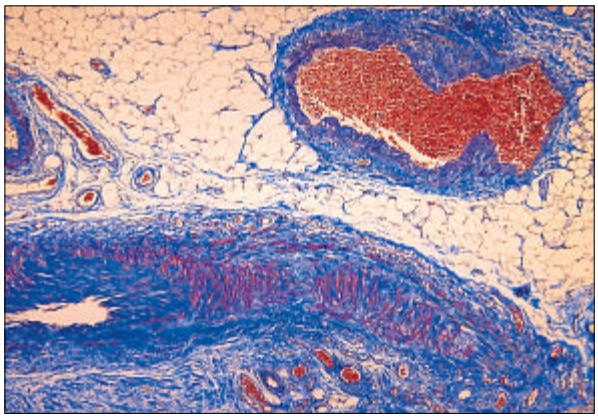

Fig. 3. The smooth muscle bundles that surrounded the blood vessels stained red for Masson trichrome stain (Masson Trichrome, × 100).

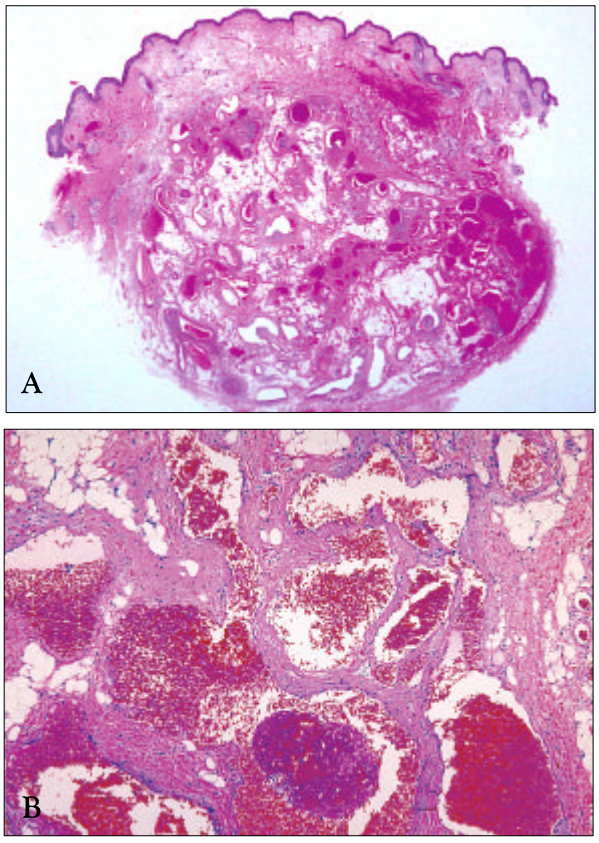

Fig. 2. (A) Low-power view showing a subcutaneous, well-circumscribed tumor (H & E, × 12.5). (B) The tumor is composed of mature adipocytes, blood vessels, and smooth muscle cells arranged in well-defined fascicles and around vessels (H & E, × 100).

cutaneous angiomyolipoma, and published a complete study of their original patient in 1991. But, Fitzpatrick et al.[6] prefer the term cutaneous angiolipoleiomyoma to cutaneous angiomyolipoma for two reasons. First, they believe that it is most closely related to cutaneous angioleiomyoma and adding a descriptive syllable to acknowledge the lipomatous component is accurate from a descriptive standpoint. Second, the use of the latter term may cause confusion with the renal angiomyolipoma. However, later, several cases by other authors were reported under the term angiomyolipoma[2-4,7].

Clinically, cutaneous angiomyolipoma presents as a solitary, asymptomatic nodule on the acral skin and the ear, that is usually mistaken for a cyst or lipoma. It arises later in life and is not associated with tuberous sclerosis or renal angiomyolipoma. In our case, there was no known personal or familial history of tuberous sclerosis. It is characterized histologically by mature adipocytes, thick-walled blood vessels, and smooth muscle cells arranged in well-defined fascicles and around blood vessels. Masson trichrome stain will separate the muscular from fibrous components that are invariably present. The smooth muscle cells have a more mature fascicular arrangement than in the renal analog, and stain negatively for HMB-45, in contrast to renal angiomyolipoma[8,9]. We also investigated HMB-45 reactivity using a commercially available monoclonal antibody, and there was no reactivity.

The histologic differential diagnosis includes angiolipomas, arteriovenous hemangiomas, subcutane-

ous cavernous hemangiomas, and angioleiomyoma. From a histogenetic viewpoint, cutaneous angiomyolipoma is most closely related to the more common angioleiomyoma and arguably could be considered as an angioleiomyoma with fatty replacement[6,8]. Indeed Hachisuga et al.[10] performed a clinicopathologic study on 562 cases of angioleiomyomas. They noted the presence of small groups of mature fat cells in 16 cases and suggested that these tumors might represent hamartomas. But, other authors believe these two disease are different[5,6]. Clinically, angioleiomyoma is often a painful nodule occuring chiefly on the lower extremities of middle aged women, but angiomyolipoma is more common in men and appear as painless nodules on the ear or acral skin. Histologically, an angioleiomyoma rarely has any collections of lipocytes, and elastic tissue stains usually fail to demonstrate elastic laminae.

Most cases of angiolipoleiomyoma have been successfully treated with simple excision, and there has been no reported case of recurrence in the dermatologic literature.

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