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Papular Angioplasia

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Papular angioplasia is a benign vascular tumor characterized by asymptomatic red papules on scalp or face. A 65-year-old woman had a dome shaped red papule on her forehead for 10 years. Histopathologic findings of the lesion showed vascular proliferations composed of atypical endothelial cells, some of which were protruded into vascular lumina or arranged in two to three layers. Fibroblast-like or histiocyte-like cells were scattered around the vessels in the dermis. Both endothelial cells and fibroblast-like or histiocyte-like cells were stained with anti-factor VIII-related antigen antibody. (Ajou Med J 1999; 4(1): 69~71)

Key Words: *Papular angioplasia, Endothelial cells, Anti-factor VIII-related antigen*

INTRODUCTION

The term "papular angioplasia" suggested in 1970 by Wilson-Jones and Marks[1] is a pseudomalignant tumor arising from vascular tissues. Papular angioplasia is characterized by multiple soft, reddish papules measuring a few millimeters in diameter on the face or scalp. It may involute spontaneously. Histopathologically atypical vascular proliferations are seen in the dermis. The capillary lumina are lined by large, protruding, columnar endothelial cells,[2] which in part form a double layer.[3] The stroma between the vessels contains numerous pleomorphic fibroblast or histiocyte-like cells. Many of them appear atypical, being hyperchromatic and occasionally multinucleated.[1] Extravasated erythrocytes may be found in the stroma.[3]

We report a case of papular angioplasia that appears as a single lesion on forehead, and identify the atypical cells by immunohistochemical staining.

CASE REPORT

A 64-year-old woman visited our clinic for the evaluation of a lesion on her forehead that had developed 10 years prior to the visit. There was neither change in the characteristics of the lesion since its development nor subjective symptom. There were no histories of drug intake, trauma or prior skin lesions in that area. Physical examination showed a pea sized reddish colored, dome shaped, soft papule on the forehead (Fig. 1). The lesion was completely excised.

Histopathologic examination of the biopsy specimen revealed thin and thick walled vascular proliferations in the upper and mid dermis (Fig. 2). Mild lymphohistiocytic infiltrations were shown in the connective tissue, especially near skin appendages. Many erythrocytes were seen in the vascular lumina. The endothelial cells of the proliferated vessels were large round, ovoid, cuboidal or occasionally atypical shaped (Fig. 3), and sometimes protruded into the vascular lumina or arranged in two to three layers. There were histiocyte-like or fibroblast-like cells in the connective tissue between the vessels. The cells had variable shapes and sizes (Fig. 3). On immunohistochemical studies, anti-factor VIII-related antigen antibody was strongly stained to the endothelial cells of thin and thick walled blood vessels. The histiocyte-like or fibroblast-like cells in the stroma were also positively stained with anti-factor VIII-related antigen antibody (Fig. 4). Anti-actin antibody was usually positively stained to smooth muscle cells of the vessel walls, but was not stained to the endothelial

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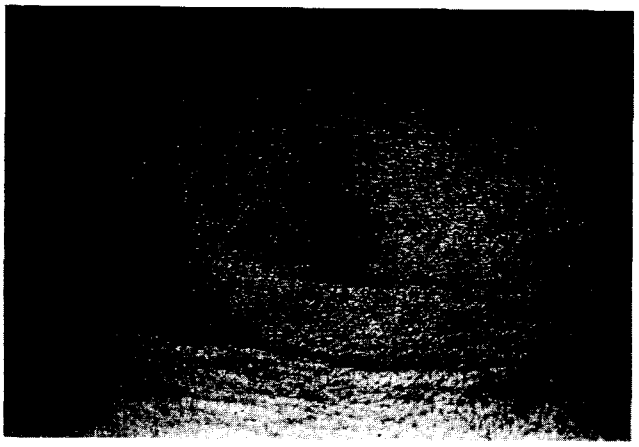

Fig. 1. A pea sized reddish colored, dome shaped, soft papule was on the forehead.

Fig. 2. Thin and thick walled vascular proliferations were seen in the dermis (H&E, ×40).

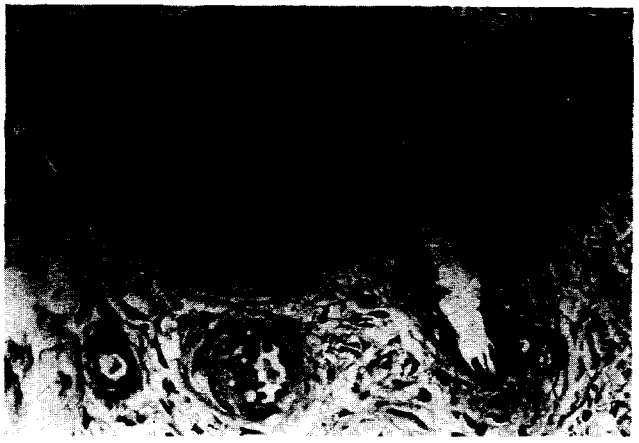

Fig. 3. The endothelial cells of the vessels were large round, ovoid, cuboidal or atypical shaped, which sometimes protruded into the vascular lumina (▲). The histiocyte-like or fibroblast-like cells in the connective tissue had variable shapes and sizes (H&E, ×100).

Fig. 4. The endothelial cells of the vessels and the histiocyte-like or fibroblast-like cells (▲) in the stroma were positively stained with anti-factor VIII-related antigen antibody (Immunoperoxidase staining, ×100).

cells of the vessels.

DISCUSSION

Papular angioplasia is a benign vascular tumor consisting of proliferations of abnormal vessels and atypical, fibroblast or histiocyte-like cells in the dermis.[1] The atypical and pleomorphic endothelial cells make histologically malignant appearances. Before the term papular angioplasia was suggested, some authors reported cases of atypical vascular lesions compatible with papular angioplasia as Kaposi's sarcoma[2] and atypical pyogenic granulomas.[3]

Wilson-Jones and Marks[1] described the components of the dermal infiltrates in papular angioplasia as abnormal capillaries and atypical, pleomorphic connective tissue cells, but the "connective tissue cells" were not exactly defined. Peterson and colleages[3] described that many of the stromal cells appeared to be arising from the endothelium, retained the same appearance as that of endothelial cells lining the blood vessels, and there was a gradual transition of these cells into elongated forms, apparantly fibroblasts. The atypical endothelial cells are different from normal endothelial cells ultrastructurally and histochemically. The atypical endothelial cells closely resemble histiocytes, but they are negatively stained for lysozyme,[4] and are positively stained for factor VIII-

related antigen antibody.[4,5] In an another word they have a propensity for endothelial cell differentiation.

Because of their morphologic resemblance to histiocyte, the atypical endothelial cells, have been often referred to histiocytoid endothelial cells.[6] There are some diseases that share histologic features characterized by distinctive histiocytoid endothelial cells, including angiolymphoid hyperplasia with eosinophilia, arteriovenous hemangioma, and papular angioplasia.[6~9]

Angiolymphoid hyperplasia with eosinophilia shows a single or multiple papules less than 1 cm in diameter on face and scalp.[10] Histologically, it shows proliferations of blood vessels lined by histiocytoid endothelial cells which form two to three layers in parts.[11] The inflammatory infiltrates are different from papular angioplasia in that they contain eosinophils[12] and sometimes plasma cells and mast cells,[13] but atypical fibroblast or histiocyte-like cells are absent. Eosinophils are increased also in the peripheral blood of the patients.[11]

Arteriovenous hemangioma shows a solitary, dark red, asymptomatic papule on head or extremities. Histologically, densely aggregated vessels are observed in the dermis, and erythrocytes and thrombi are occasionally found in the vascular lumina.[14] It is different from papular angioplasia in that there are much more vascular lumina, less intervascular stroma, and less individual endothelial cells are seen in arteriovenous hemangioma. Extravasated erythrocytes and hemosiderins are also found in the dermis.[10]

Angioendotheliomas represent red brown patches to purpule red papules or nodules.[15] Histologically they show dilatated vessels with proliferation of endothelial cells that often fill the vascular lumina with fibrin thrombi, and display mild cytologic atypia.[10]

Hemangioendotheliomas also show proliferation of endothelial cells, but the degree of endothelial cell proliferation is far greater, and solid cords of endothelial cells are found. Clinically, they usually show infiltrative and progressive growth and metastases,[3] and have a tendency to recur after an incomplete excision.[13]

In our case, a histopathologic finding is compatible with papular angioplasia, and a positive staining with factor VIII-related antigen established the fibroblast or histiocyte-like cells in the dermis to be endothelial cells.

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