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Solitary Angiokeratoma Developed in one Area of Lymphangioma Circumscriptum

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We describe a boy who developed a dark brown colored nodule in an area of lymphangioma circumscriptum following repeated local injuries. The nodule showed the clinical and pathologic features of solitary angiokeratoma. It is possible that previous injuries predisposed the lesion to the development of solitary angiokeratoma.

Key Words: *Solitary angiokeratoma, lymphangioma circumscriptum*

INTRODUCTION

Solitary angiokeratoma is an independent entity, which has been categorized as the 5th entity of angiokeratoma (Imperial and Helwig, 1967). Solitary angiokeratoma is thought to be caused by various factors such as injury or chronic irritation as an injury-response of the papillary vessel wall (Imperial and Helwig, 1967). We experienced a case of solitary angiokeratoma, seen on one area of lymphangioma circumscriptum. In this paper, the case is presented with a review of the previous literature.

CASE REPORT

A 15-year-old boy came to the Department of Dermatology of Hanyang University Hospital because of a dark brown colored nodule and vesicular lesions on the right flank. Since his early childhood, he had these vesicular lesions. A dark brown colored nodule developed in an area of the lesions 3 years before his visit, and there was a history of local injuries.

On physical examination, this dark brown colored keratotic nodule was surrounded by clear, tense, vesicular lesions on the right flank (Fig. 1). Laboratory examinations including complete blood cell counts, erythrocyte sedimentation rate, routine urinalysis, chest roentgenogram, and liver function test were normal. The lesion was surgically excised. Microscopic exami-

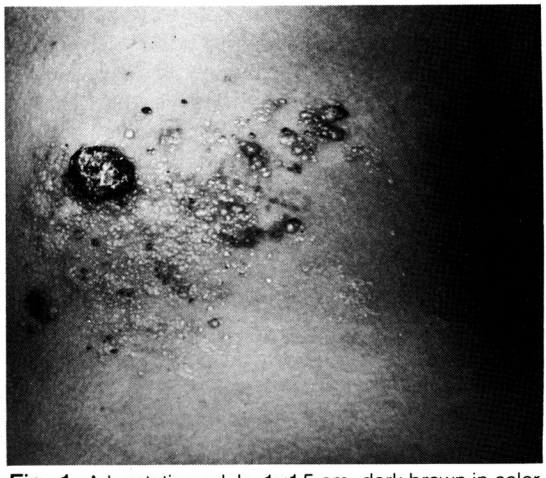

Fig. 1. A keratotic nodule, 1×1.5 cm, dark brown in color with grouped vesicular lesions on the right flank.

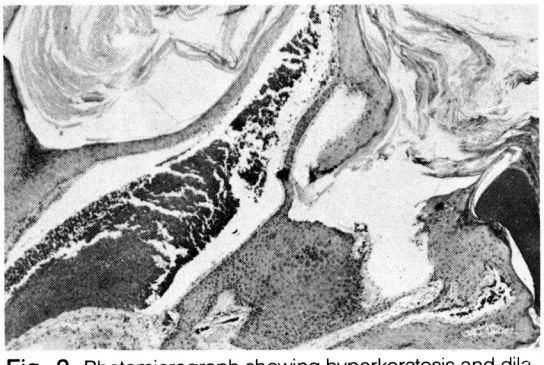

Fig. 2. Photomicrograph showing hyperkeratosis and dilatation of blood vessels containing many red blood cells, which are surrounded by elongated rete ridges. (H & E, ×40)

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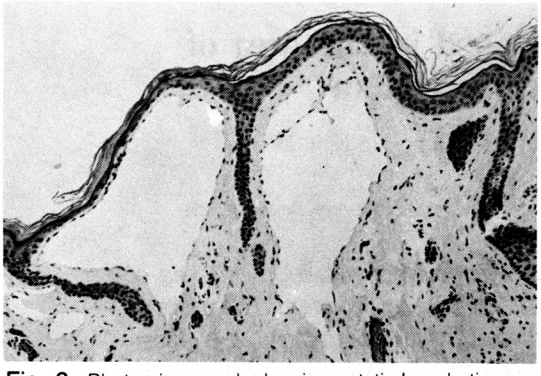

Fig. 3. Photomicrograph showing ectatic lymphatic vessels in the papillary dermis, with overlying thinning of stratum malpighii. (H & E, ×100)

nation of the nodule showed greatly dilated capillaries containing erythrocytes just beneath, or enclosed within, the papillomatous epidermis with hyperkeratosis and irregular acanthosis (Fig. 2). In the vesicular lesions, markedly dilated lymph vessels lined by only a few endothelial cells in the upper dermis and hyperkeratotic epidermis were present (Fig. 3).

DISCUSSION

Solitary and multiple angiokeratomas were proposed as the 5th category of angiokeratoma, following angiokeratoma corporis diffusum, angiokeratoma Mibelli, angiokeratoma Fordyce, and angiokeratoma circumscriptum. Clinically, solitary and multiple angiokeratomas often resemble malignant melanoma in appearance, but should be differentiated from nevocellular nevus, seborrheic keratosis, pigmented basal cell carcinoma, hemangioma and capillary aneurysm (Hayen, 1966; Loria et al., 1958; Epstein et al., 1965). Histopathologically, solitary and multiple angiokeratomas reveal hyperkeratosis, acanthosis, vascular dilatation of the papillary vessels, and a close relation between the epidermis and vascular lacunae. This case was thought to be a typical solitary angiokeratoma because of the clinical as well as histopathological features. It was suggested that solitary and multiple angiokeratomas occur as a response to various factors such as injury and chronic irritation to the wall of the papillary vessels (Imperial and Helwig, 1967). It is reasonable to assume that, in this case also, repeated injuries served as trigger for the development of angiokeratoma (Goldman et al., 1981). Athough it is possible that the solitary angiokeratoma and lymphangioma circumscriptum arose independent of each other in our patient, it may be that this angiokeratoma was formed in the underlying lymphangioma circumscriptum by the repeated injuries. In addition to the case reported in this article, we have recently seen solitary angiokeratoma in a patient with pretibial epidermolkysis bullosa (Maeda, 1987). To our knowledge, our case is the first reported instance of solitary angiokeratoma in an area of lymphangioma circumscriptum.

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