

A Case of Acquired Cutaneous Smooth Muscle Hamartoma

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Smooth muscle hamartoma was first described by Stokes in 1923 as the disease characterized by increase of well-defined bundles of smooth muscle fibers in the dermis. It can be either congenital or acquired. Acquired ones are often in association with Becker's nevus. We present a case of acquired smooth muscle hamartoma that did not show any pigmentation or hair growth. (*Ann Dermatol* 14(3) 161~163, 2002).

Key Words : Becker's nevus, Smooth muscle hamartoma, Acquired

In human skin, smooth muscle is present in 3 locations: 1) the arrectores pilorum of the hair follicle, 2) the scrotal and labial dartos muscle as well as smooth muscle within the nipple and areola, and 3) that within the walls of arteries and veins¹. Cutaneous smooth muscle hamartomas are benign proliferation of smooth muscle bundles within the dermis. They are either congenital or acquired. Acquired ones are usually accompanied by increased hairs and pigments. There have been two cases of smooth muscle hamartoma associated with Becker's nevus^{2,3} and one case of congenital smooth muscle hamartoma⁴ in Korea. Those two cases associated with Becker's nevus both showed hyperpigmentation, and one of them also showed hypertrichosis on the lesion. We describe an acquired smooth muscle hamartoma that does not show hyperpigmentation or increased hairs.

CASE REPORT

A 57 year-old woman visited our department with skin lesion on her right neck area. The lesion was flesh colored, palm sized, papular surfaced, in-

durated plaque that had developed since 14 years old (Fig. 1). There was no hyperpigmentation or hypertrichosis. A pseudo-Darier's sign could not be elicited. Laboratory examinations were all negative or within normal limit except blood sugar with FBS 197mg/dl and pp2hr 286mg/dl. Biopsy was performed on her right neck. Tissue sections were stained with hematoxylin and eosin and Masson's trichrome. Analysis of tissue section revealed small muscles in the reticular dermis (Fig. 2). The individual cells composing the bundles were spindle-shaped and possessed cigar-shaped nuclei. Masson's trichrome stain of the tissue section confirmed the presence of increased smooth muscle fibers (Fig. 3).

DISCUSSION

Smooth muscle hamartoma was first described by Stokes et al in 1923. Most of them are congenital, and its frequency found to be more common than initially believed with 1:1000 to 1:27000 live birth⁵. Congenital smooth muscle hamartomas are often covered by prominent vellus hairs. It probably represents aberrant development of pilar smooth muscle (arrectores pilorum) during fetal maturation⁵. Acquired ones have been reported most frequently in association with a Becker nevus⁶.

The typical clinical feature of smooth muscle hamartoma shows slightly hyperpigmented patch or plaque, usually located on the trunk, arms, legs, or buttocks. Rarely, it is described as a patch with

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Fig. 1. Flesh colored, palm sized, papular surfaced, indurated plaque on the right neck area.

Fig. 3. The bundles of muscle fibers are readily distinguishable from collagen bundles by the Masson's trichrome stain. (Masson's trichrome stain, $\times 400$).

perifollicular papules without prominent hair⁵. Usually, lesions show hyperpigmentation and hypertrichosis. Some can produce wormlike movements ("vermiculation") and stroking may induce transient induration with piloerection (pseudo-Darier's sign)⁷. Histologic findings show increased bundles of smooth muscle fibers in the dermis, not attached to the hair follicles. The bundles of muscle fibers are readily distinguishable from collagen bundles by the trichrome stain.

Since the clinical features of acquired smooth muscle hamartoma are similar to Becker's nevus, it has been called congenital Becker's nevus. It has also been suggested that congenital smooth muscle hamartoma could be at one end of a spectrum of

Fig. 2. Increased bundles of smooth muscle fibers in the dermis. The individual cells composing the bundles are spindle-shaped and possess cigar-shaped nuclei. (H&E, $\times 400$).

dermal smooth muscle proliferative disorders, and that Becker's melanosis could represent the entity at the opposite end⁸. This hypothesis is supported by some cases that show the features intermediate between smooth muscle hamartoma and Becker's nevus^{9,10}. Recently, some authors suggested that it is probably appropriate to describe these lesions as distinct disease entities, each with a spectrum of hamartomatous growth that involves the epidermis (hyperpigmentation), hair follicle (hypertrichosis), and dermis (smooth muscle)⁶. There are more diseases to differentiate from smooth muscle hamartoma other than Becker's nevus. They are piloleiomyoma, congenital pigmented hairy nevocellular nevus, and solitary mastocytoma¹¹. Piloleiomyoma clinically present as a painful or tender discrete nodules. Solitary mastocytoma shows Darier's sign and histologically mast cells under Toluidine blue or Giemsa stain. Congenital pigmented hairy nevocellular nevus does not show induration and there is no smooth muscle hyperplasia histologically.

The clinical diagnosis of smooth muscle hamartoma remains difficult due to the lack of specific diagnostic criteria. It should be suspected in any congenital hairy lesions and biopsy should be performed to confirm the diagnosis. However, since there is no evidence of malignant transformation of these lesions nor the evidence of an association with any other malformation, treatment is not necessary except for cosmetic purposes.

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