

CASE REPORT

A Case of Acquired Smooth Muscle Hamartoma on the Sole

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A smooth muscle hamartoma is a benign proliferation of smooth muscle bundles within the dermis. It arises from smooth muscle cells that are located in arrector pili muscles, dartos muscles, vascular smooth muscles, muscularis mamillae and the areolae. Acquired smooth muscle hamartoma (ASMH) is rare, with only 10 such cases having been reported in the English medical literature to date. Most of these cases of ASMH were shown to have originated from arrector pili and dartos muscles. Only one case was reported to have originated from vascular smooth muscle cells. A 21 year-old woman presented with a tender pigmented nodule, with numbness, on the sole of her foot, and this lesion had developed over the previous 18 months. The lesion showed no hyperpigmentation or hypertrichosis, and the biopsies demonstrated increased smooth muscle bundles in the dermis, and especially around the blood vessels. Moreover, the specimens stained positive with Masson trichrome stain and α -smooth-muscle actin antibodies, thus supporting our diagnosis of ASMH of the foot sole. Herein, we report on a rare case of ASMH on the foot sole, and this lesion originated from vascular smooth muscle cells. This type of case has not been previously described in the English medical literature.

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-Keywords-

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INTRODUCTION

Smooth muscle hamartomas (SMH) are a result of benign proliferation of smooth muscle bundles in the dermis. SMH-associated smooth muscle cells originate from arrector pili-, dartos-, vulvar-, mamillary- and vascular wall-muscles¹⁻³. SMH is characterized by slightly pigmented plaques that contain vellus hairs, and SMH is subdivided into two types: congenital smooth muscle hamartoma (CSMH) and acquired smooth muscle hamartoma (ASMH)¹⁻³. ASMH is a very rare form of SMH that was first described by Wong and Solomon¹ in 1985, with only such 10 cases having been reported in the English medical literature (Table 1). These lesions usually occur on the proximal extremities^{1,2}, scrotum^{4,7}, vulva⁸, shoulders² and trunk⁹⁻¹¹.

CASE REPORT

A 21 year-old women presented with an 18-month history of a slow-growing 0.5×0.5 cm nodule on the right sole (Fig. 1). She complained that the lesion was mildly tender

Table 1. The reported cases of acquired smooth muscle hamartoma in the English medical literature

Case	Gender/Age	Onset	Origin
1	M/28	14	Arrector pili muscle
2	M/19	12	Arrector pili muscle
3	M/60	—	Dartos muscle
4	M/33	25	Dartos muscle
5	M/65	64	Dartos muscle
6	M/24	23	Dartos muscle
7	F/36	31	Dartos muscle
8	M/18	16	Arrector pili muscle
9	M/35	25	Arrector pili muscle
10	F/57	14	Arrector pili muscle
Present case	F/21	19	Smooth muscle cells of blood vessels

and somewhat numb. Any evidence of trauma, allergic reaction, hyperpigmentation and hypertrichosis was not found and the Pseudo-Darier's sign could not be elicited. Moreover, the results of the complete blood count, blood chemistry and urinalysis were all within the normal limits. A skin biopsy specimen from the lesion revealed bundles of smooth muscle cells with cigar shaped nuclei, and these cells were arranged haphazardly throughout the dermis and especially around the blood vessels (Fig. 2). Furthermore, the spindle cells stained positive with Masson trichrome stain and α -smooth-muscle actin antibodies. The nodule was subsequently excised, and there has been no evidence of recurrence.



Fig. 1. A skin colored, 0.5x0.5 cm sized tender nodule on the right sole.

DISCUSSION

SMH is usually manifested at birth, but it can also appear later in life. CSMH is a congenital lesion that's principally composed of smooth muscle with or without vellus hair and/or epidermal hyperpigmentation. It is usually located on the trunk and extremities and it appears as variable sized papules, patches or plaques⁴. Transient piloerection or elevation of a lesion that's induced by rubbing is referred to as Pseudo-Darier's sign and this is the characteristic diagnostic clue for CSMH¹⁻⁴. ASMH appears later in life as a slightly hyperpigmented or hypertrichotic patch or plaque with associated itching, pain or numbness. These lesions are usually negative for Pseudo-Darier's sign⁷. The histopathologic hallmark of SMH is a local increase of large, randomly oriented bundles of smooth muscle cells with central, cigar-shaped nuclei. These smooth muscle bundles are arranged haphazardly around blood vessels in the dermis, and the smooth muscle bundles stain positive with Masson trichrome stain and α -smooth-muscle actin antibodies (Fig. 3).

The differential diagnosis for SMH includes leiomyomas, angioleiomyomas and Becker's nevus. In contrast to SMH, the smooth muscle bundles in leiomyomas are more tightly interlacing and poorly demarcated, and they can also be painful or tender. Angioleiomyomas contain thick-walled blood vessels with smooth muscle fibers deposited in concentric layers that merge with the muscular stroma. Hence, angioleiomyomas can be differentiated from SMH by the histological findings.

Becker's nevus often resembles SMH both clinically and histologically. Becker's nevus is a well-defined, uniformly hyperpigmented plaque with hypertrichosis. Histologically, Becker's nevus exhibits an abnormal number of large, deeply seated terminal hair follicles that are sometimes associated with hyperplastic pilar smooth muscles.

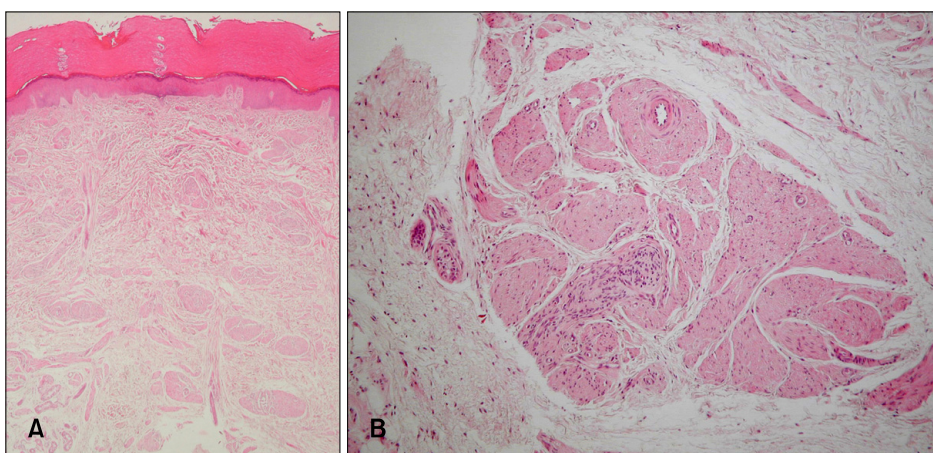


Fig. 2. An increased number of haphazardly oriented bundles of smooth muscle cells were observed in the dermis (A: H&E stain, $\times 40$; B: H&E stain, $\times 100$).

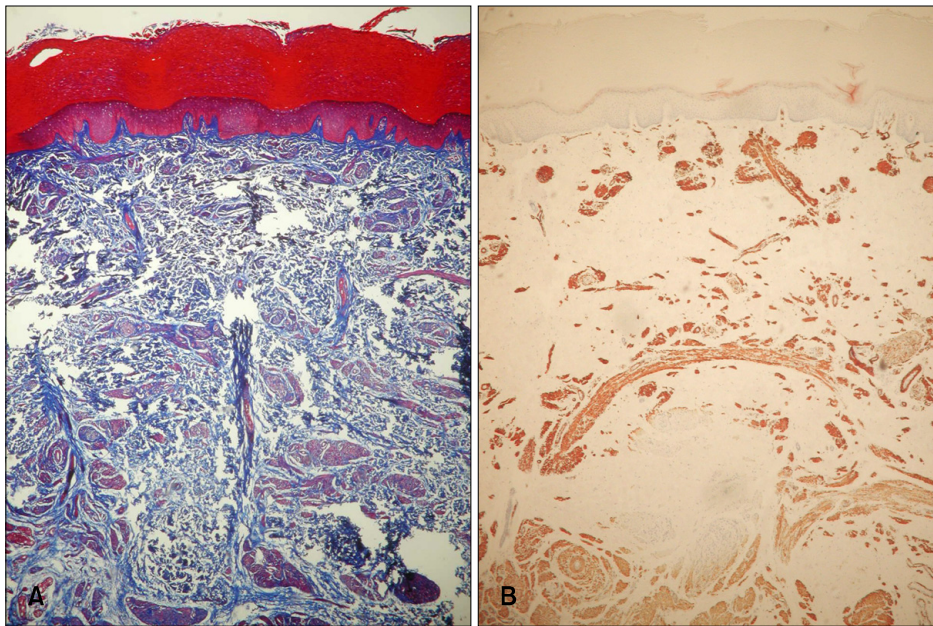


Fig. 3. The haphazardly arranged smooth muscle bundles were mainly located around blood vessels (A: Masson's trichrome stain, $\times 40$, B: α -smooth-muscle actin antibody stain, $\times 40$).

However, Becker's nevus can also present with a proliferation of dermal smooth muscle. Because of the overlapping histological features, some authors have speculated that Becker's nevus and SMH are two polar entities at either end of a continuous spectrum^{2,4,9}. Since Wong and Solomon¹ reported on the first case of ASMH in 1983, only 10 cases of ASMH have currently been reported in the English medical literature. ASMH is usually located on the skin of the forearm, chest, neck, scrotum, penis, major labium or shoulder. Five cases of ASMH had been reported to originate from the dartos muscle and 5 cases have been reported to originate from the arrector pili muscle (Table 1). In this case, we hypothesize that the hamartomatous proliferation of smooth muscle cells originated from blood vessel walls due to the haphazard arrangement of smooth muscle bundles around the dermal vasculature and the fact that arrector pili and dartos muscles are not normally present in foot soles. To the best of our knowledge, this is the first case of a smooth muscle hamartoma of the sole to be reported in the English medical literature.

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