A Case of Multiple Acquired Depressed Smooth Muscle Hamartoma

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Key Clinical Message

This case report presents a 40-year-old male with multiple acquired depressed smooth muscle hamartoma. The report for the first time describes smooth muscle hamartoma characterized by multiple distribution, acquired onset, and depressed appearance.

Introduction

Smooth muscle hamartoma (SMH) is a benign skin tumor characterized by the proliferation of smooth muscle bundles, primarily originating from the pilomotor muscles. SMH most commonly presents in the lumbar and sacral regions and typically manifests within the first six months of life. The clinical presentation of SMH is characterized by pale brown macules with slightly indistinct borders. To date, there have been few reported cases of multiple SMH in the literature [1]. Additionally, there are very limited reports of acquired SMH[2]. In this report, we present a unique case of acquired SMH with multiple distribution and depressed appearance.

Case Report

A 40-year-old French male patient was referred to us with a 7-year history of multiple pigmented macules on the trunk and extremities. The patient had no history of diabetes mellitus or injections, including steroids and insulin. No relevant clinical manifestations were observed among his family members. Physical examination

revealed multiple, slightly depressed, and pigmented macules ranging from 1 to 3 cm in diameter without hypertrichosis on the trunk and extremities (**Figure 1**).

Histopathological examination demonstrated irregularly distributed, bundled pale eosinophilic structures of various sizes with oval-shaped nuclei in the upper dermis by hematoxylin and eosin staining (**Figure 2a**). There was no discernible increase in the number of melanocytes in the epidermis or hair follicles. Masson trichrome staining showed that the irregularly distributed and variously sized bundled structures were less stained than collagen fibers in the dermis (**Figure 2b**). These findings collectively indicated an aberrant proliferation of mature smooth muscle in the upper dermis. Based on these findings, the diagnosis of SMH was made. The patient was followed for 6 months, during which time there was no change in the cutaneous manifestations.

Discussion

This case presents a distinct clinical manifestation of SMH in three notable aspects: its multiple occurrences, its acquired nature, and its depressed appearance. Initially, a unique appearance of multiple occurrences in the case should be reviewed. Because of several case reports with multiple Becker's nevi [3,4] , in the current case, Becker's nevi were considered as a potential differential diagnosis. However, due to the absence of hypertrichosis and the lack of epidermal changes, the present case is reasonably diagnosed as SMH[5]. Based on our current understanding, there have been a few case reports of multiple SMH in localized areas of the body, such as the head and scrotum [6,7] . Nevertheless, no instances of widespread multiple SMH throughout the body have been documented in the English literature. Considering it, our case is deemed to be exceedingly rare.

While a higher incidence of single lesions in congenital instances has been suggested [1], a definitive clinical presentation of acquired cases has not been fully established, primarily due to the limited number of case reports regarding acquired instances. The scarcity of clinical photographs in the literature might lead to the difficulty in identifying cases with similar clinical presentations to this case. Although there is a report of multiple SMH arising in a single family [8,9] , this case had no evidence of familial occurrence.

The onset of this case at 36 years of age is particularly noteworthy. In general, SMH is considered congenital, and acquired cases are relatively rare. According to a case report published in 2021 [2], there had been only 25 cases of acquired SMH reported internationally. Among these, 14 cases occurred after the age of 30. In light of these findings, it is conceivable that the presented case is not uncommon as an acquired instance of SMH.

Lastly, a distinctly depressed appearance exhibited in the present case should be described. The case should be clinically distinguished from anetoderma, atrophoderma of Pasini and Pierini, scleroderma en plaques, lupus profundus, and discoid lupus erythematosus. In our case, we did not observe the disappearance or fragmentation of elastic fibers as is typically seen in anetoderma [10]. Additionally, there was a remarkable proliferation of smooth muscle, which is not usually seen in atrophoderma of Pasini and Pierini [11,12] . Furthermore, lilac rings indicative of scleroderma en plaques, subcutaneous indurations associated with lupus profundus, or erythema with hyper- or hypo-keratosis suggestive of discoid lupus erythematosus were all absent. These considerations reinforce the diagnosis of SMH in this case. As per our current understanding, there exists only a solitary report documenting SMH with a depressed appearance[13], making our case exceptionally uncommon.

In conclusion, we have encountered a case of SMH with distinctive features. We consider this case to be clinically rare for three reasons: its multiple occurrences, its acquired nature, and its depressed appearance. It is necessary for dermatologists to consider SMH as one of the differential diagnoses when patients show multiple depressed macules.

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Statement of Ethics: The study protocol was approved by The Ethics Committee of The Jikei University School of Medicine and the patient provided written informed consent.

Consent statement: Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

Data Availability Statement: Additional data sharing is not applicable to this article due to ethical restrictions.

Author Contributions

Kaoru Chiba: The author contributed to data curation, resources, and drafting the original paper.

Itaru Dekio: The author contributed to resources and preparation for paper writing.

Isami Uno: The author contributed to resources.

Yoshimasa Nobeyama: The author contributed to conceptualization and project administration.

Akihiko Asahina: The author contributed to review and supervision.

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Figure legends

Figure 1. Clinical manifestation

Multiple depressed and pigmented macules, ranging in diameter from 1 to 3 cm, are observed on the back.

Figure 2. Histopathological findings

a) Irregularly distributed and variously sized bundled pale eosinophilic structures with oval-shaped nuclei are present in the upper dermis (hematoxylin and eosin stain, ×100). b) Irregularly distributed and variously sized bundled structures were less stained than collagen fibers in the dermis by Masson trichrome stain (×100).