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Cold-induced cutaneous smooth muscle hamartoma

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Abstract

Cutaneous smooth muscle hamartomas (SMHs) are benign proliferations of smooth muscle tissue, typically congenital and characterized by hyperpigmentation, hypertrichosis, and a positive pseudo-Darier's sign. We report an unusual case of an early-onset acquired SMH in a 49-year-old woman. The lesion first appeared during adolescence and remained asymptomatic and clinically inapparent except upon exposure to cold. Cold stimulation induced a transient, linear, wrinkled, hyperpigmented papular lesion on the left thigh, which resolved spontaneously within seconds after rewarming. Histological examination revealed dermal fibrosis and well-organized lobules of smooth muscle cells, confirming the diagnosis of SMH without requiring immunohistochemical analysis. This case highlights a unique and transient presentation of SMH revealed exclusively by cold exposure and reversible with rewarming. It broadens the clinical spectrum of SMHs and the importance of external stimuli in their diagnosis.

Introduction

In human skin, smooth muscle is predominantly associated with hair follicles, forming the arrector pili muscle,[1] and is also present in the walls of blood vessels. Smooth muscle hamartomas (SMHs) are benign tumors typically presenting as asymptomatic growths in various parts of the body, including the skin, gastrointestinal, respiratory, and genitourinary tracts.[2] Although not malignant, these hamartomas can be associated with complications especially if they are large, and can compress surrounding structures, causing pain or even dysfunction of the affected organs.

Classically, SMHs are congenital, but can sometimes appear or may be discovered during childhood or adulthood. In the skin, these benign tumors typically present as follicular papules or indurated plaques with irregular contours, sometimes pigmented or associated with hypertrichosis, generally fixed, whose relief may be accentuated by friction (Darier's pseudo-sign). However, some cases of fluctuating SMHs with spontaneous appearance and disappearance have been described.[3,4]

We report an original case of an early-onset acquired SMH, fluctuating, and revealed exclusively by cold stimulation.

Case Report

A 49-year-old woman, without relevant medical history, presented with a solitary lesion localized on the anterior and lateral faces of the left thigh, which was noticed during adolescence. The specificity of this lesion was that it only appeared when exposed to cold, being completely invisible the rest of the time. It was not hives, but rather a linear, hyperpigmented lesion that rapidly developed a wrinkled appearance after exposure to cold. The lesion spontaneously resolved within a few seconds upon

warming or gentle friction of the skin (Figures 1 and 2). No hypertrichosis was observed. The lesion was neither painful nor pruritic. Histopathological examination of a biopsy described a fibrous dermis with a proliferation of smooth muscle cells arranged in regularly distributed lobules, enabling us to make the diagnosis of SMH (Figure 3).

Discussion

Cutaneous SMHs are benign lesions, characterized by disorganized smooth muscle tissue. Congenital SMHs are the most described in the literature, present at birth and typically diagnosed in early childhood, as unilateral cutaneous plaques or nodules, predominantly located on the trunk or lower limbs, classically hyperpigmented and associated with hypertrichosis.[5] In contrast, acquired SMHs develop later in life and may arise secondary to trauma, chronic inflammation, or other environmental factors.[6] Histologically, acquired and congenital SMHs are similar, characterized by well-differentiated smooth muscle cells arranged in a disorganized manner, often forming irregular fascicles and bundles. Given the absence of histological differences, the clinical history and circumstances of discovery are the key elements for diagnosis.

Alternative diagnoses include cutaneous leiomyoma, dermatofibroma, glomus tumor, and myofibroma. These differential diagnoses may have similar clinical presentations, but are not fluctuating. Histological examination is essential to establish a definitive diagnosis. To ensure the diagnosis, SMHs can be distinguished from other mesenchymal tumors through specific immunohistochemical markers, including smooth muscle actin and desmin, which are highly expressed in smooth muscle cells.[7] This was not the case for our patient whose lesion presented typical architecture and smooth muscle cells on anatomopathological analysis.

A notable feature of acquired forms is the possible revelation or accentuation of the lesion by stimuli such as cold exposure. This phenomenon could be explained by a reflex contraction of smooth muscle fibers, making the lesion temporarily more visible or palpable, which is not generally observed in congenital forms. Thus, the revelation of a SMH by cold is a clinical characteristic to be aware of, particularly in acquired forms.

Conclusions

This case of SMH is original because of its early-onset acquired nature in adolescence, and diagnosed in adulthood, but above all because of its mode of revelation by cold, rapidly reversible after rewarming, allowing us to expand the semiology of this benign tumor.

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Figure 1. A) Before cold exposure; **B**) after cold exposure with a bag of ice rubbed few seconds on both legs, the SMH appears as a linear papular patch.

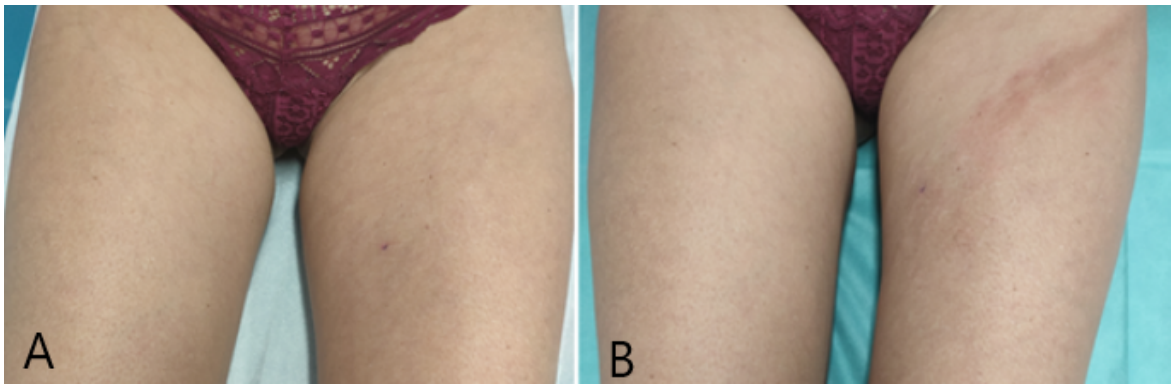

Figure 2. Zoom of the area showing the linear, papular, wrinkled lesion, corresponding to the smooth muscle hamartoma highlighted by cold exposure.

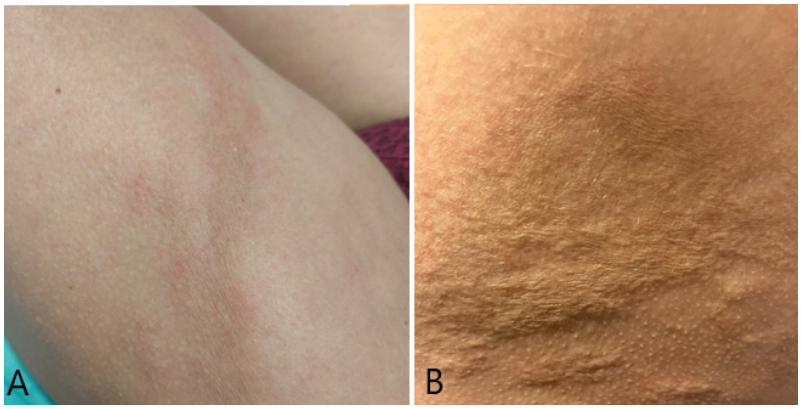

Figure 3. Histological examination shows: **A**) hematoxylin–eosin staining (HES) × 1; **B**) HES × 5; **C**) epidermal lining of preserved thickness without significant abnormalities; **D**) dermal fibrosis; and **E**) proliferation of smooth muscle cells. No further analysis, including immunohistochemistry, was required, based on the typical histological appearance of smooth muscle cells.