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CLINICAL CASE

Acquired smooth muscle hamartoma with sebaceous component

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Abstract

Background: *Acquired smooth muscle hamartoma (ASMH) is a rare benign lesion characterized clinically by hyperpigmented plaques with hypertrichosis and some follicular papules. The main histologic finding is the presence of disorganized smooth muscle bundles in the dermis. Only 25 cases of ASMH have been reported in the literature.* **Clinical case:** *We present the case of an 18-year-old male who reported a pigmented area and increased hair growth on the left hemifacial with one year of evolution. Clinically, a plaque was observed in the preauricular region and on the left cheek with a linear Blaschkoid path, consisting of hyperpigmentation, hypertrichosis, and some papular lesions, with negative pseudo-Darier sign. Histological analysis showed an increase in the number of smooth muscle bundles in the middle and deep dermis surrounding abundant sebaceous glands and numerous hair follicles in different stages of evolution.* **Conclusions:** *The sebaceous component in this lesion was prominent. Therefore, we considered this lesion part of a spectrum where the acquired smooth muscle hamartoma and folliculosebaceous cystic hamartoma are found at the extremes. This case would fall in the middle of the range, as it combines both histological features.*

Keywords: *Acquired smooth muscle hamartoma. Sebaceous. Folliculo-sebaceous.*

Hamartoma de músculo liso adquirido con componente sebáceo

Resumen

Introducción: *El hamartoma de músculo liso adquirido (HMLA) es una lesión benigna adquirida, poco frecuente, caracterizada clínicamente por presentar placas hiperpigmentadas, con hipertricosis y algunas pápulas foliculares. El principal hallazgo histológico es la presencia de abundantes haces de músculo liso desorganizados en la dermis. Se han reportado solo 25 casos de HMLA en la literatura.* **Caso clínico:** *Se presenta el caso de un paciente de sexo masculino de 18 años que refirió una zona pigmentada y el aumento de vello en la hemicara izquierda con un año de evolución. Clínicamente se observó una placa en la región preauricular y mejilla izquierda con trayecto lineal blaschkoide, constituida por hiperpigmentación, hipertricosis y algunas lesiones papulares, con signo pseudo-Darier negativo. Histológicamente se encontró un aumento en el número de haces de músculo liso en la dermis media y profunda rodeando abundantes glándulas sebáceas, así como numerosos folículos pilosos en diferentes estadios de evolución.* **Conclusiones:** *El componente sebáceo en esta lesión fue muy marcado, por lo que se considera que forma parte de un espectro donde en los extremos se encuentran el*

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hamartoma de músculo liso adquirido y el hamartoma quístico folículo sebáceo. El presente caso se encontraría en medio, ya que combina ambas características histológicas.

Palabras clave: *Hamartoma de músculo liso adquirido. Sebáceo. Folículo-sebáceo.*

Introduction

Smooth muscle hamartomas (SMH) are rare benign neoformations characterized by increased density of smooth muscle bundles in the dermis and are divided into congenital and acquired. Congenital smooth muscle hamartomas (CSMH) are the most common and present as plaques with hyperpigmentation and hypertrichosis. Conversely, acquired smooth muscle hamartoma (ASMH) is a rare condition with a heterogeneous clinical presentation[1]. The following is the first case of acquired smooth muscle hamartoma with a sebaceous component.

Clinical case

We describe the case of an 18-year-old male patient with no relevant history of the current condition, who came for consultation due to a pigmented area with excess hair on the left cheek. According to the patient, this condition was uncomfortable because it forced him to shave that area more frequently compared to the contralateral side of the face. The dermatosis had been progressing for a year with no further symptoms. A physician prescribed topical steroids and emollients, but no improvement was observed.

Physical examination revealed a localized dermatosis on the left hemiface affecting the preauricular region, which distributed caudally following the Blaschko lines of the face, passing through the mandibular ramus and gonial angle until reaching the left cheek. The dermatosis consisted of irregular, poorly defined, 2 x 8 cm, a hyperpigmented, grayish-brown plaque with abundant short, coarse hair on its surface and 1-2 mm follicular papules (Figure 1). No piloerection, erythema, or induration of the plaque was observed when the lesion was rubbed (negative pseudo-Darier sign). The evolution had been asymptomatic up to that time.

The patient underwent two skin biopsies: the first from the left cheek and the second from the left preauricular region. In the first biopsy, orthokeratosis was found covering the epidermis with discrete acanthosis and hyperpigmentation of the basal layer. In addition, a fragment with isolated smooth muscle fiber bundles was identified in the mid-dermis, with no involvement of any other structure at the epidermis or dermis level (Figure 2). The second biopsy showed orthokeratosis with discrete and regular acanthosis and hyperpigmentation of the basal layer with numerous follicular sebaceous structures. Sebaceous glands of different sizes were observed from the superficial dermis to the deep dermis. Miniaturized follicular outgrowths were observed in the follicular component, and numerous intermingled smooth muscle bundles in the external, mid, and deep dermis with collagen disruption and sebaceous glands surroundings. Masson's trichrome staining was positive, showing enhancement of muscle bundles in red over dermal collagen tissue in blue (Figure 3). Based on the clinicopathologic correlation, acquired smooth muscle hamartoma with a sebaceous component was diagnosed. Treatment with 2% hydroquinone was administered for 3 months, which partially improved hyperpigmentation.

Discussion

The word hamartoma comes from the Greek word *hamartomein* (to fail, error). Hamartomas are benign neoplastic lesions that combine several cellular elements of various origins. Cutaneous hamartomas are tumor-like malformations in which there is overgrowth and a defective mixture of normal skin components[1,2].

A smooth muscle hamartoma (SMH) is a benign proliferation of smooth muscle cells within the dermis. Smooth muscle cells of the skin can be found in the erector pili muscle, scrotal musculature (dartos), vulvar musculature, nipple or areola (*muscularis mamillae*), and dermal blood vessel wall[3,4]. Histologically, smooth muscle cells are spindle-shaped with a central "cigar-shaped" nucleus and eosinophilic cytoplasm without striations[1].

Smooth muscle hamartomas can be divided into two types: congenital and acquired. To date, the reported cases of congenital hamartomas showed their origin in the erector pili muscles, and those acquired, in the erector pili mucles, as well as in dartos, vulvar, or dermis blood vessel muscles[3-5].

Congenital smooth muscle hamartoma (CSMH) is the most frequent form; the first case was described in 1969 by Sourreril et al.[6]. Clinically, it presents as a hyperpigmented plaque with hypertrichosis and some

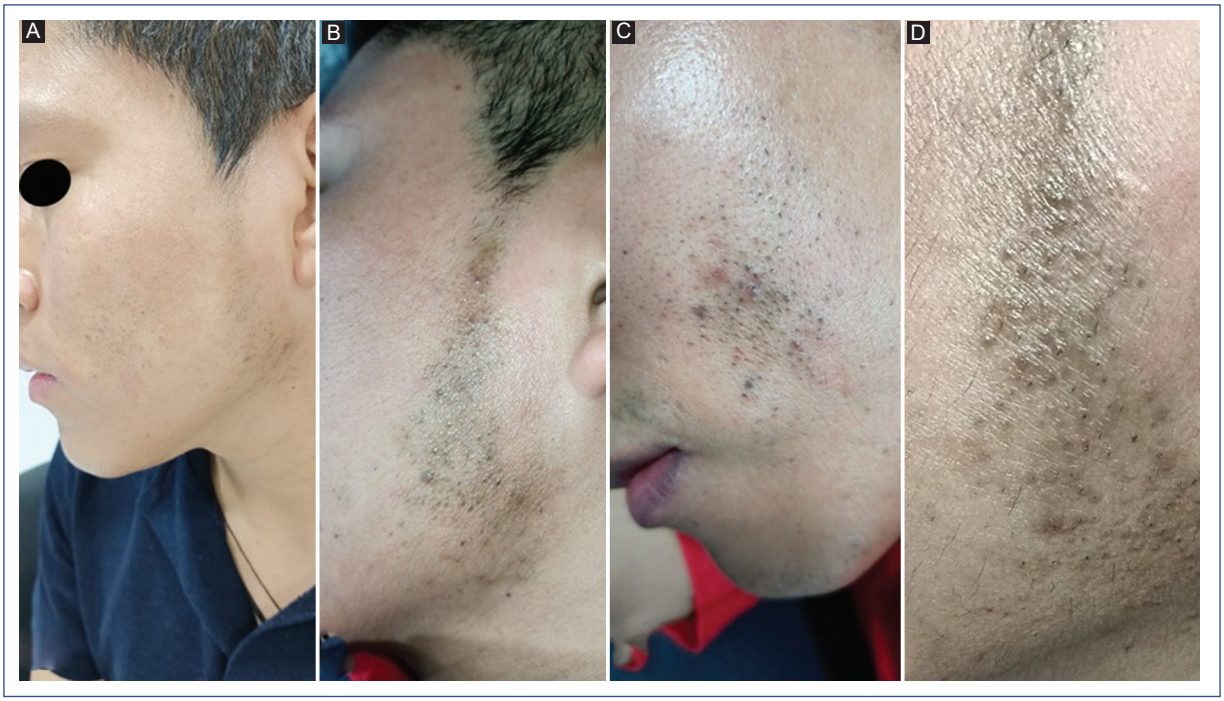

Figure 1. A: plaque following a Blaschkoid distribution on the left hemiface. **B-C:** the shape of the plaque is irregular, with poorly defined borders in the preauricular region and on the left cheek. **D:** close-up shows grayish-brown hyperpigmentation, an abundance of coarse, short hairs, and millimetric papular lesions.

papular elements located predominantly on the trunk or extremities. The positive pseudo-Darier sign is considered typical of the congenital form. This sign consists of the triggering transient edema, induration, or piloerection upon rubbing the lesion[1,7]. This sign represents a neural component of the hamartoma, not an actual histamine release, as seen in the true Darier sign in cutaneous mastocytosis[8].

Acquired smooth muscle hamartoma (ASMH) is a rare variety first described by Wong and Solomon in 1985[5]. This form appears later in life compared to the congenital variety. According to a literature search, only 25 cases of ASMH have been described (Table 1).

ASMH also presents clinically as plaques with hyperpigmentation and a variable degree of hypertrichosis, as well as small papules on the surface. However, very heterogeneous morphologies have been described, ranging from nodular lesions to large neoformations, forming masses up to 30 cm in size[9,10]. The topographies reported are the scalp[10], face[11], neck[12,13], trunk[9,14-18], upper extremities[5,19,20], lower extremities[3,21,22], vulva[23,24], and penoscrotal area[4,25-30]. The present case is the second case of facial localization reported worldwide. De Alba et al. (2001) reported the first case of ASMH located on the face of an 18-year-old male patient, with a hyperpigmented plaque with thick, short, and abundant hair, with perifollicular hemispherical lesions affecting the right cheek. Histologically, the deep dermis showed numerous smooth muscle bundles and hair follicles in different evolutionary stages[11].

ASMH is usually asymptomatic, although some patients present with pruritus, pain, or paresthesia[3,23]. A case with hyperhidrosis in the lesion was recently reported[22]. The acquired variant usually shows a negative pseudo-Darier sign[10]. On histopathology, long bundles of smooth muscle cells are randomly arranged at different dermis levels, and muscle cells have a central "cigar-shaped" nucleus[23]. Smooth muscle bundles are positive in Masson's trichrome stain. By immunohistochemistry, alpha-actin and desmin positivity confirm the muscular histologic origin[14]. The age of presentation of the reported cases varies, ranging from 18 to 70 years, with no gender predominance.

Regarding the differential diagnosis of smooth muscle hamartoma, dermatologists should consider leiomyomas, angioleiomyomas, Becker's nevus, and connective tissue nevi.

Leiomyomas are single or multiple neoformations of papular appearance, smooth, red, brown, or skin-colored. They adhere to the skin but not to deeper tissues and

Table 1. Summary of cases of acquired smooth muscle hamartoma reported in the international literature

Author/Country	Sex (age)	Time of evolution	Topography	Morphology	Pigmentation	Hypertrichosis	Pseudo-Darier	Symptoms	Location/Origin
Wong and Solomon, 1985 (Country not reported)[5]	Male (28 years)	14 years	Forearm	Solitary plaque of 2-3 cm	Yes	No	NR	Asymptomatic	Dermis and hypodermis, erector pili muscle
Darling et al., 1993 (USA)[12]	Male (53 years)	10 years	Neck	Erythematous, indurated plaque	No	No	Negative	Asymptomatic	Dermis, erector pili muscle
De la Espriella et al., 1993 (France)[14]	Female (18 years)	3 years	Right breast	15 cm plaque composed of papules	NR	NR	NR	Asymptomatic	Dermis, erector pili muscle
Hsiao and Chen, 1995 (Taiwan)[25]	Male (33 years)	8 years	Scrotum	Multiple skin-colored papular and nodular lesions	No	No	Negative	Asymptomatic	Dermis, dartos muscle
Semerci et al., 1997 (Turkey)[26]	Male (65 years)	8 months	Penoscrotal region	Thickening and edema	No	No	NR	NR	Dermis, dartos muscle
Quinn and Young, 1997 (USA)[27]	Male (60 years)	Several months	Scrotum	2.8 x 1.8 cm nodular neoformation	No	No	NR	Intermittent purulent drainage	Dermis, dartos muscle
Kwon et al., 2000 (Korea)[23]	Female (30 years)	5 years	Vulva (labia majora)	10 x 4 cm yellowish indurated plaque with multiple 2-5 mm papules	No	No	NR	Pruritus	Dermis, dartos labialis muscle
De Alba et al., 2001 (Mexico)[11]	Male (18 years)	3 years	Face (left cheek)	10 x 5.5 cm plaque with 1-2 mm perifollicular hemispheric lesions	Yes	Yes	Negative	Asymptomatic	Dermis, erector pili muscle
Ryu et al., 2002 (Korea)[13]	Female (57 years)	43 years	Neck (right side)	Indurated plaque with skin-colored papular surface	No	No	Negative	NR	Dermis, erector pili muscle
Morales-Callaghan et al., 2005 (Spain)[9]	Male (18 years)	2 years	Chest (left side)	Triangular plaque with skin-colored papules and hypertrichosis	No	Yes	Positive	Asymptomatic	Dermis, erector pili muscle
Oiso et al., 2005 (Japan)[4]	Male (24 years)	6 months	Scrotum	Diffuse thickening and edema with skin-colored papules	No	No	Negative	Asymptomatic	Dermis, dartos muscle
Bari and Rahman, 2006 (Pakistan)[15]	Male (19 years)	7 years	Back and left shoulder	Red-brown indurated plaque, irregular surface, with mottled pigmentation	No	No	Positive	Pruritus	Dermis, erector pili muscle
Zarineh et al., 2008 (USA)[16]	Male (49 years)	NR	Back (upper left portion)	Nodular pinkish neoformation 1.2 cm with grayish pigmentation in the center	Yes	No	NR	NR	Dermis, erector pili muscle. Two components: melanocyte cords intermingled with smooth muscle bundles
Lee et al., 2009 (Korea)[3]	Female (21 years)	18 months	Right sole	Nodular neoformation 0.5x0.5 cm	No	No	Negative	Paresthesia	Dermis, vascular smooth muscle

(Continues...)

Table 1. Summary of cases of acquired smooth muscle hamartoma reported in the international literature *(Continued)*

Author/Country	Sex (age)	Time of evolution	Topography	Morphology	Pigmentation	Hypertrichosis	Pseudo-Darier	Symptoms	Location/Origin
Yancovitz et al., 2009 (USA)[19]	Female (52 years)	46 years	Left arm	Indurated plaque of 15 cm and erythematous papules on its surface.	Yes	Yes	Negative	Pruritus	Dermis, erector pili muscle
Monteagudo et al., 2010 (Spain)[17]	Male (32 years)	26 years	Back and right shoulder	Macular neoformation with dark brown punctiform spots and a 2 cm plaque at one end of the lesion	Yes	Yes	Positive	Asymptomatic	Dermis, erector pili muscle. Two components: lentiginous melanocytic hyperplasia and smooth muscle bundles
Toeima et al., 2010 (England)[24]	Female (55 years)	NR	Vulva	Lichenification	No	No	NR	Vaginal xerosis	Dermis, dartos labialis muscle
Matsuda et al., 2011 (Japan)[20]	Male (22 years)	16 years	Upper right arm	Dark brown, indurated plaque 8x10 cm with follicular papules on its surface	Yes	No	NR	Pruritus, Meyerson's phenomenon (eczematization)	Dermis, erector pili muscle
Adulkar et al., 2014 (India)[18]	Male (32 years)	6 months	Back (left side)	Plaque with multiple hyperpigmented follicular papules	NR	Yes	NR	Asymptomatic	Dermis, erector pili muscle
Chen et al., 2015 (Taiwan)[28]	Male (58 years)	10 years	Scrotum	A large mass of 30 cm composed of multiple skin-colored, firm papular, and nodular neoformations	No	No	NR	Gait disturbances, erectile dysfunction	NR
Bogetti et al., 2016 (Italy)[29]	Male (70 years)	1 year	Scrotum	A large mass of 20 x 32 cm, with multiple papular neoformations on its surface erythema and ulceration	NR	NR	NR	Inability to stand upright	Dermis, dartos muscle
Desai et al., 2017 (India)[21]	Female (28 years)	8 months	Right mid-heel	Erythematous plaque	No	No	Negative	Asymptomatic	Dermis and hypodermis, erector pili muscle
Jain et al., 2018 (India)[10]	Male (25 years)	6 years	Scalp skin	Multiple indurated, hyperpigmented, poorly defined plaques. The most extensive, measuring 4.5 x 6 cm. Wrinkled skin appearance	Yes	No	Negative	Asymptomatic	Dermis, erector pili muscle
Ladha and Remington, 2019 (Canada)[22]	Female (29 years)	6 years	Left leg	A 7 cm plaque, circular sclera, firm, with a conglomerate of skin-colored papules, warty appearance, with perilesional hyperpigmentation	Yes	No	Negative	Pain and hyperhidrosis	Dermis, erector pili muscle
Chen et al., 2019 (Taiwan)[30]	Male (41 years)	1 year	Penis (dorsal side)	Erythematous indurated plaque	No	No	Positive	Pruritus	Dermis, dartos muscle

NR: not reported.

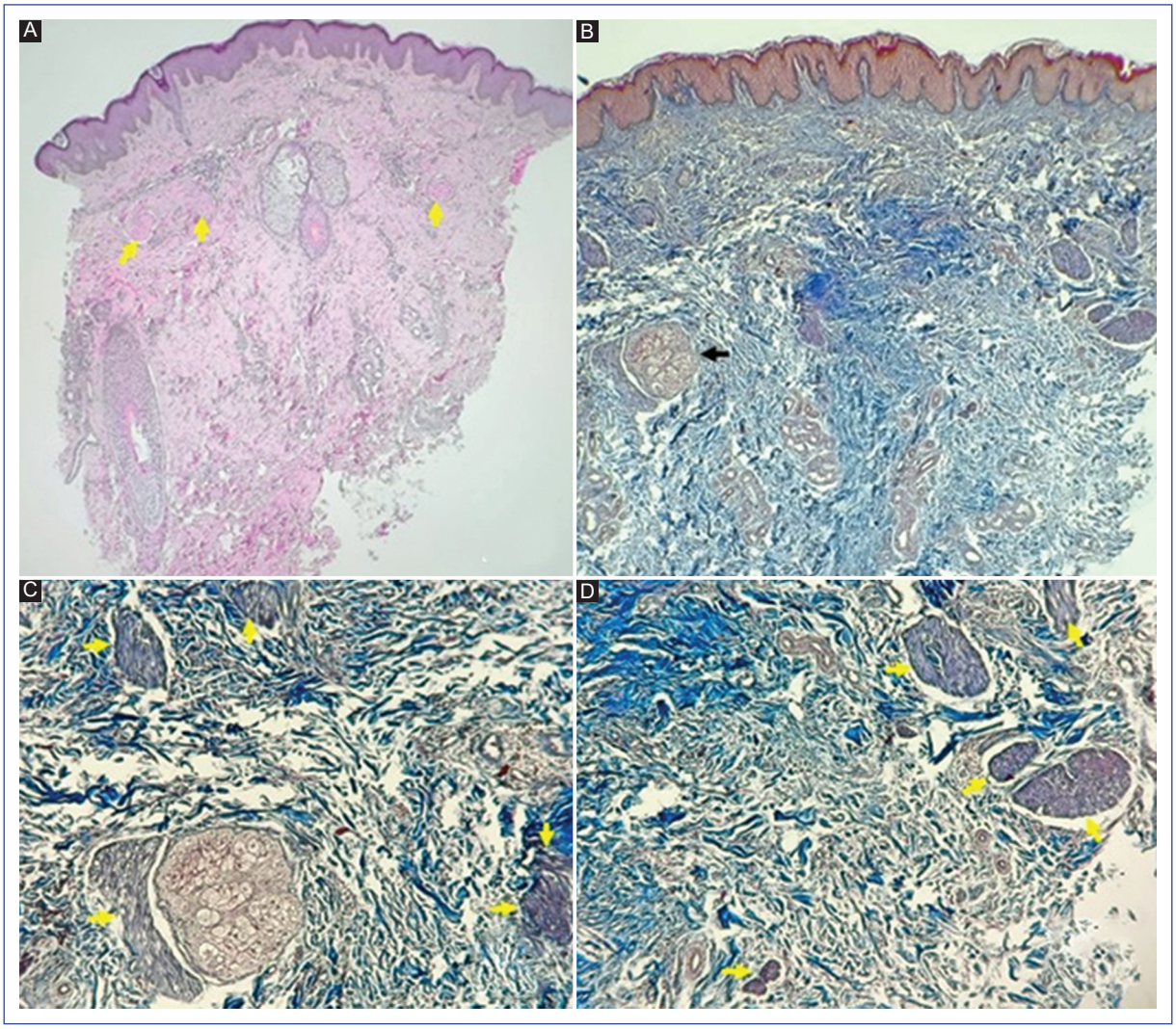

Figure 2. Histopathology of left cheek lesion. **A:** numerous smooth muscle bundles (arrows) can be observed in the mid and deep dermis. Presence of a small adnexa in the mid dermis and perivascular inflammatory infiltrate (hematoxylin-eosin stain, 4x). **B:** multiple isolated smooth muscle bundles are observed, one surrounding the sebaceous gland (arrow) with no sebaceous follicle continuity (Masson's trichrome stain, 10x). **C:** close-up of the smooth muscle bundles in the collagen surrounding the sebaceous gland (arrows) (Masson's trichrome stain, 40x). **D:** close-up of the smooth muscle bundles (Masson's trichrome stain, 40x).

are painful to palpation. They predominate on the extensor surfaces of the extremities, followed by the trunk, head, and neck. On histology, they contain tightly intertwined and poorly demarcated smooth muscle bundles.

Angioleiomyomas are single neoformations of papular or nodular appearance. These lesions are red or brown, well-demarcated, slow-growing, and painful on palpation. They mainly affect the lower extremities and, less frequently, the upper extremities. Angioleiomyomas contain thick-walled blood vessels with smooth muscle fibers arranged in concentric layers.

Becker's nevi are mixed-shape, light brown, hyperpigmented spots with fine hair on the surface. The presentation of these lesions can be multiple and bilateral, sometimes with no hypertrichosis. They are located on the face, shoulders, and anterior chest. Clinically and histologically, Becker's nevus is the most similar entity to smooth muscle hamartoma. It has an increased number of large, deeply seated hair follicles that are sometimes associated with hyperplastic dermal smooth muscle. Due to the overlapping histologic features, some authors consider Becker's nevus and smooth muscle hamartoma to be polar conditions at either end of a spectrum[3,9,15,31]. ASMH has been associated with other neoformations such as melanocytic nevi[16] or nevus spilus[17].

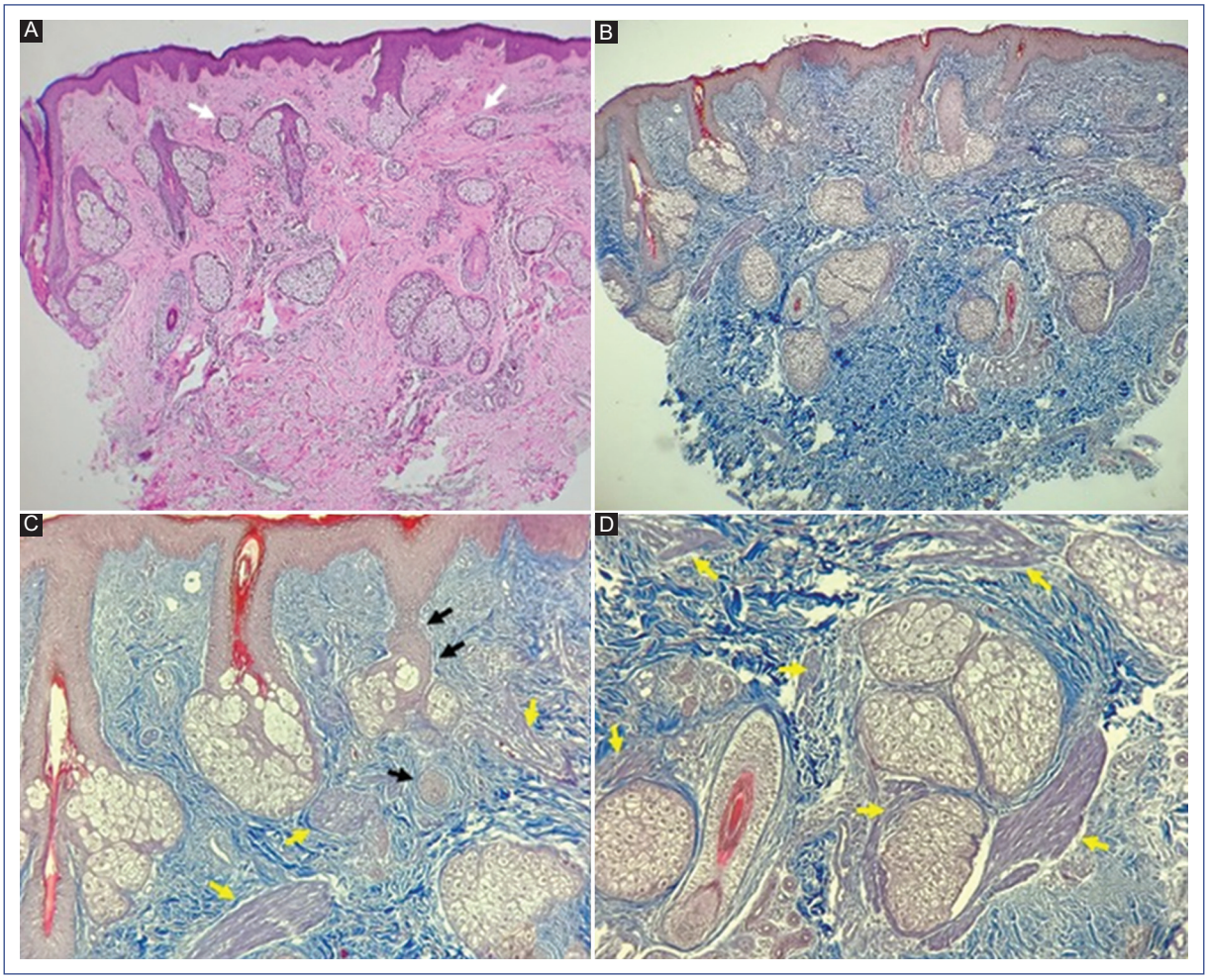

Figure 3. Histopathology of the left preauricular lesion. **A:** panoramic view showing numerous sebaceous glands, some miniaturized (arrows), and smooth muscle bundles (hematoxylin-eosin stain, 4x). **B:** abundant smooth muscle bundles embedded in the collagen bundles and surrounding the mature sebaceous glands (Masson's trichrome stain, 10x). **C**: abundant smooth muscle bundles in the collagen (yellow arrows); miniaturized sebaceous follicles (black arrows) (Masson's trichrome stain, 20x). **D:** smooth muscle bundles (arrows) surrounding a mature sebaceous gland (Masson's trichrome stain, 40x).

Another diagnosis to consider is connective tissue nevi. These neoformations can be congenital or acquired, single or multiple. Clinically, their appearance is papular or nodular, yellowish or skin-colored with a mamelon-shaped surface. The collagenous forms of these nevi are characterized by thickened collagen bundles in the reticular dermis, whereas elastic tissue nevi show thickened elastic fibers in the reticular dermis[32].

Given the benign nature of ASMH, treatment is unnecessary; however, it can be surgically removed for cosmetic reasons, or lasers can be used to remove excess hair[1] and reduce epidermal pigmentation. Some patients improve with a combination of several types of laser [pulsed light, Nd: YAG (neodymium-doped yttrium aluminum garnet), alexandrite, diode, among others] used similarly to how they are used for Becker's nevus. Wavelengths range from 504 nm to 10,600 nm[33].

The case reported here is considered atypical because of the clinical manifestations and histology. The patient showed numerous smooth muscle bundles arranged randomly, with an orientation unrelated to hair follicles and interspersed with abundant sebaceous glands throughout the thickness of the dermis. The patient also showed a significant number of hair follicles in different stages of evolution.

According to the literature, acquired smooth muscle hamartoma is derived from and constituted by smooth muscle, whereas folliculosebaceous cystic hamartoma presents cystic structures characterized by aberrant proliferation of hair follicles, sebaceous glands, and

blood vessels surrounded by fibrous septa and a large number of sebocytes scattered throughout the thickness of the dermis[34].

Clinically, both hamartomas present similar findings, such as nodular and xanthochromic plaque-like lesions and other pigmentations. In this regard, two cases reported were found, one as a multiple-plaque smooth muscle hamartoma[10] and the other reported as the first case of folliculosebaceous cystic hamartoma with smooth muscle component[35]. In both cases, many smooth muscle bundles with numerous sebaceous glands of various sizes were identified on histopathologic imaging. Therefore, the patient presented here could be an intermediate form of hamartoma within a spectrum in which, at one end, there is a pure smooth muscle hamartoma and, at the other, a folliculosebaceous cystic hamartoma.

We propose to name this variant smooth muscle hamartoma with a sebaceous component, which has not been previously described to the best of our knowledge.

The present case is unusual because it is a case of an acquired smooth muscle hamartoma with a unique topography, such as the face, and mixed histopathologic findings. This article emphasizes the clinical and histopathological variability associated with both acquired smooth muscle hamartoma and folliculosebaceous cystic hamartoma.

Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this study.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author has this document.

Conflicts of interest

The authors declare no conflicts of interest.

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