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ORIGINAL ARTICLE

Dermoscopy of cutaneous smooth muscle neoplasms: a morphological study of 136 cases

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Abstract

Background A variety of cutaneous smooth muscle neoplasms may arise in the skin and are frequently unrecognized by clinicians. There is sparse data relating to the dermoscopy of piloleiomyomas (PL), and nothing has been published about the dermoscopy of angioleiomyomas (AL) and leiomyosarcomas (LS).

Objectives To evaluate the morphological findings of a large series of cutaneous PL, AL and LS under dermoscopic observation, comparing these findings among them.

Methods Digital dermoscopic images of 136 histopathologically confirmed cases of cutaneous smooth muscle neoplasms (114 PL, 13 AL and 9 LS) collected from 10 Hospitals in Spain, Austria and Italy were evaluated for the presence of dermoscopic structures and patterns.

Results The pattern composed of a symmetric, total delicate pigment network with the variable presence of multiple hypopigmented areas in a painful lesion is the most common dermoscopic pattern associated with PL. This pattern was found in 69.3% of PL and in no cases of AL and LS. The most common and characteristic pattern associated with AL was the one composed of symmetric pink-reddish tumour with vessels, white structures and the absence of ulceration, which was found in 46.2% of AL, but also in 3.5% of PL, and in 22.2% of LS. Finally, the most common pattern associated with LS was the one composed of an asymmetric, multilobulated tumour with linear-irregular or polymorphic-atypical vessels and white structures, which was found in 44.4% of cases, but also in 0.9% of PL and in 15.4% of AL.

Conclusion Dermoscopy is helpful in improving the diagnostic accuracy of PL. The dermoscopic patterns associated with AL and LS were more variable and less specific.

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Conflicts of interest

None declared.

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Introduction

Smooth muscle is found in the skin in three different locations: arrector pili muscles, blood vessels walls and specialized muscle of genital skin, which includes scrotum, vulva and nipple. A variety of cutaneous muscle neoplasms may arise in the skin.[1–3]

Cutaneous leiomyomas are uncommon benign smooth muscle neoplasms (BSMT) of skin which are classified into three types according to the site of origin in: piloleiomyomas (PL) (derived from the arrector pili muscle), angioleiomyomas (AL) (from vascular smooth muscle) and genital leiomyomas (from

smooth muscle of the genital skin).[1,2] PL can be solitary or multiple (most commonly) and present as firm, reddish-brown or skin-coloured papules or nodules located on the trunk (multiple lesions) or extremities (solitary lesions).[1,2] Clinically, AL typically present as solitary, subcutaneous or dermal firm nodules located most frequently on the lower extremities.[1] Approximately 90% of PL and 50% of AL are painful.[1–3] Hereditary leiomyomatosis and renal cell cancer syndrome (HLRCC), also known as Reed syndrome, is a rare genetic disorder caused by a germline heterozygous mutation of the fumarate hydratase tumour suppressor gene, that predisposes individuals to multiple cutaneous PL, renal cell carcinomas and, in women, uterine leiomyomas.[2]

Superficial leiomyosarcoma is a rare soft tissue neoplasm of smooth muscle derivation which comprises approximately 2%–3% of all soft tissue sarcomas and can be subdivided into cutaneous or subcutaneous forms with markedly different clinical and prognostic implications.[3] The cutaneous leiomyosarcomas (LS) are believed to derive from the arrector pili or genital dartoic muscle, whereas the subcutaneous type is thought to arise from the smooth muscle wall of blood vessels. LS classically present as a solitary, firm, skin-coloured to red brown nodules or plaques. These tumours typically range from 0.3 to 4 cm in diameter and most commonly occur on the extremities.[3]

Cutaneous muscle neoplasms are uncommon tumours which are frequently unrecognized and misdiagnosed by clinicians. Dermoscopy is a non-invasive technique, which has greatly improved the diagnostic accuracy of pigmented and non-pigmented skin tumours. We consider it worthwhile to communicate the dermoscopic features of a large series of smooth muscle tumours that could improve its clinical diagnostic accuracy.

Material and methods

Dermoscopic images of 114 histopathologically proven cases of PL, 13 cases of AL and nine cases of LS collected from 10 Hospitals in Spain, Austria and Italy were evaluated for the presence of dermoscopic features. Clinical data were obtained for each case, including: age and sex of patients, the anatomical location of the lesions, the presence of pain and the clinical diagnosis or differential diagnoses before excision. Dermoscopic images of each lesion were obtained using DermLite Foto (3Gen, LLC, Dana Point, CA, USA) mounted on a digital camera at 20- to 50-fold magnification. No pressure was used to avoid the collapse of the vessels in the lesions. All the lesions in this study were evaluated for the presence of dermoscopic features by two of the contributing authors (JB and PZ).

Continuous variables are presented as means and standard deviation (SD). Categorical variables were presented as frequencies and percentages. Comparisons between proportions in the different cutaneous smooth muscle neoplasms and dermoscopic features were studied using the Pearson chi-squared test and Fisher's exact probability test. For continuous variables, the differences between means were calculated performing Student's *t*-test. The *P*-value <0.05 was considered statistically significant. Statistical analysis was performed using SPSS, version 24.0 (IBM SPSS Statistics, Armonk, NY, USA).

Results

A total of 136 smooth muscle neoplasms (114 PL, 13 AL and 9 LS) from 51 patients (10 with HLRCC syndrome or Reed syndrome) were included. Mean age was 50.1 ± 15.1 years in the BSMT group (48.2 ± 15.9 years in the group of PL and 54.1 ± 12.8 years in the group of AL), and 69.6 ± 14.2 years in the group of LS (Student *t*-test, $P = 0.001$). The male-to-female ratio was 0.9 for the BSMT group and 8 for the LS group

Table 1 Clinical characteristics observed in 136 smooth muscle tumours

Clinical features		PL (%) *n* = 114	AL (%) *n* = 13	*P*1	BSMT (%) *n* = 127	LS (%) *n* = 9	*P*2
Gender†	Male	14 (48.3)	6 (46.2)	0.899*	20 (47.6)	8 (88.9)	0.031**
	Female	15 (51.7)	7 (53.8)		22 (52.4)	1 (11.1)	
Location	Head/Neck	4 (3.5)	2 (15.4)	–	6 (4.7)	5 (55.6)	–
	Trunk	27 (23.7)	1 (7.7)		28 (22)	0	
	Limbs	42 (36.8)	10 (76.9)		52 (40.9)	4 (44.4)	
	≥2 areas	41 (36)	0		41 (32.3)	0	
Pain (presence)		89 (78.1)	3 (23.1)	<0.001**	92 (72.4)	0	<0.001**
Preoperative diagnosis	Correct	85 (74.6)	0	–	85 (65.9)	0	–
	OBT	26 (22.8)	5 (38.5)		31 (24.4)	1 (11.1)	
	OMT	3 (2.6)	8 (61.5)		11 (8.7)	9 (88.9)	

*Chi-squared test, **Fisher's exact test.
†Gender of the patients (*n* = 51).
(–) *P*-value cannot be calculated because more than 20 per cent of the cells have expected frequencies of 5 or more.
AL, Angioleiomyomas; BSMT, Benign smooth muscle tumours; LS, Leiomyosarcomas; OBT, Other benign tumours; OMT, Other malignant tumours; PL, Piloleiomyomas; *P*1, *P*-value between PL and AL; *P*2, *P*-value between BSMT and LS.

(Fisher's exact test, $P = 0.0031$). The majority of BSMT were located on extremities (83/114, 72.8%), whereas more than a half of LS were located on the head (55.6%). Most PL were painful papules (89/114, 78.1%), whereas only the minority of AL (3/13, 23.1%) and none of the LS caused pain (Fisher's exact test, $P < 0.001$). The preoperative diagnosis was predominantly correct in the group of PL (85/115, 74.6%; above all in those cases with multiple lesions) and totally incorrect in the group of AL and LS. All but one LS were diagnosed as malignant tumours (four basal cell carcinomas, two squamous cell carcinomas, one dermatofibrosarcoma protuberans, one melanoma and one dermatofibroma) (Table 1).

Comparing dermoscopically PL and AL of our study (Table 2): (i) Both lesions were predominantly symmetric; however, AL were more significantly asymmetric than PL (23.1% vs. 3.5%; $P = 0.023$); (ii) Pigment network was more common in PL (96.5% vs. 30.8%, $P < 0.001$), being delicate pigment network the only morphology in both lesions (Figs 1, 2); (iii) Vascular structures were more common in AL (15.8% vs. 69.2%; $P < 0.001$), being the unfocused arborizing telangiectasias the most frequent type in PL whereas linear-irregular and polymorphic-atypical vessels in AL (Figs 3, 4); (iv) Milky-red areas were found only in AL (0% vs. 61.5%, $P < 0.001$) (Figs 3, 4); (v) White structures were found predominantly in AL (7% vs. 92.3%, $P < 0.001$) and chrysalis were only seen in AL (Figs 3, 4); and (vi) Multifocal hypopigmented areas were only seen in PL (49.1% vs. 0%, $P < 0.001$) (Fig. 2).

Comparing dermoscopically the BSMT and LS of our study (Table 2): (i) LS were less symmetric (94.5% vs. 22.2%, $P < 0.001$) and presented more ulceration (2.4% vs. 55.6%, $P < 0.001$) and multilobulated structures (2.4% vs. 55.6%, $P < 0.001$) than BSMT (Figs 5, 6); (ii) Pigment network was more common in BSMT (89.8% vs. 55.6%, $P = 0.015$), and LS showed atypical pigment network in 40% of cases and multifocal distribution in 80% of cases; (iii) Vascular structures were observed more commonly in LS (21.3% vs. 88.9%, $P < 0.001$), and the polymorphic-atypical vessels were the most frequent type in LS (62.5%) (Fig. 6); and (iv) White structures were found more frequently in LS (15.7% vs. 88.9%, $P < 0.001$).

In our study, the most characteristic and common dermoscopic pattern associated with PL was the one composed of symmetry, total delicate pigment network with the variable presence of multiple hypopigmented areas, and the absence of vascular structures and ulceration, which was found in 69.3% of PL (Figs 1, 2) and in no cases of AL and LS. The patterns associated with AL and LS were more variable and less

Table 2 Dermoscopic characteristics observed in 136 smooth muscle tumours

Dermoscopic features		PL (%)	AL (%)	*P*1	BSMT (%)	LS (%)	*P*2
Symmetry (yes)		110 (96.5)	10 (76.9)	0.023*	120 (94.5)	2 (22.2)	<0.001*
Multilobulated tumour (yes)		1 (0.9)	2 (15.4)	0.028*	3 (2.4)	5 (55.6)	<0.001*
Pigment network (yes)		110 (96.5)	4 (30.8)	<0.001*	114 (89.8)	5 (55.6)	0.015*
Type	Delicate	110 (100)	4 (100)	–	114 (100)	3 (60)	0.001*
	Atypical	0	0	–	0	2 (40)	–
Distribution	Total	98 (89.1)	0	–	98 (86.0)	0	–
	Peripheral	8 (7.3)	4 (100)	–	12 (10.5)	1 (20)	–
	Multifocal	4 (3.6)	0	–	4 (3.5)	4 (80)	–
Vascular structures (yes)		18 (15.8)	9 (69.2)	<0.001*	27 (21.3)	8 (88.9)	<0.001*
Type	ATNF	11 (61.1)	1 (11.1)	–	12 (44.4)	1 (12.5)	–
	Dotted	4 (22.2)	2 (22.2)	–	7 (25.9)	1 (12.5)	–
	Linear irregular	0	3 (33.3)	–	3 (11.1)	1 (12.5)	–
	Polymorphic	2 (11.1)	3 (33.3)	–	5 (18.5)	5 (62.5)	
Distribution	Total	5 (27.8)	2 (22.2)	–	7 (25.9)	1 (12.5)	–
	Peripheral	2 (11.1)	0	–	2 (7.4)	1 (12.5)	–
	Central	2 (11.1)	0	–	2 (7.4)	3 (37.5)	–
	Multifocal	9 (50)	7 (77.8)	–	16 (59.3)	3 (37.5)	–
White structures (yes)		8 (7.0)	12 (92.3)	<0.001*	20 (15.7)	8 (88.9)	<0.001*
Types	Chrysalis	0	3 (25)	0.242*	17 (85.0)	3 (37.5)	0.022*
	White scar-like patch	8 (100)	9 (75)	–	3 (15.0)	5 (62.5)	–
Hypopigmented areas (yes)		56 (49.1)	0	0.001*	56 (44.1)	1 (11.1)	0.079*
Milky-red areas (yes)		0	8 (61.5)	<0.001*	8 (6.3)	5 (55.6)	<0.001*
Ulceration (yes)		2 (1.8)	1 (7.7)	0.279*	3 (2.4)	5 (55.6)	<0.001*

*Fisher's exact test.

AL, Angioleiomyomas; BSMT, Benign smooth muscle tumours; LS, Leiomyosarcomas; OBT, Other benign tumours; OMT, Other malignant tumours; PL, Piloleiomyomas; *P*1, *P*-value between PL and AL; *P*2, *P*-value between BSMT and LS.

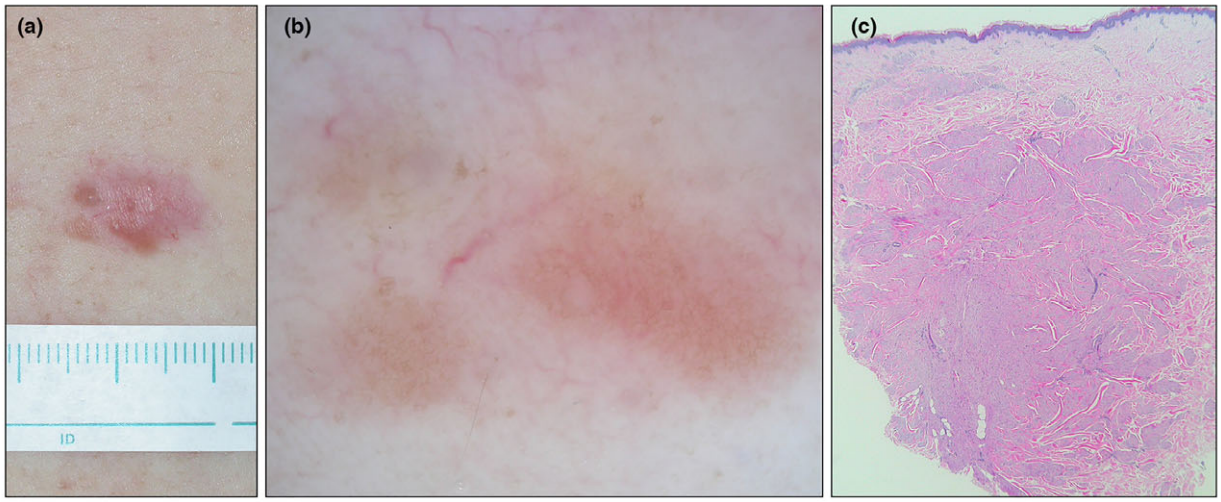

Figure 1 (a) Painful, pigmented lesion located on the trunk of a 45-year-old man with leiomyomatosis and renal cell cancer syndrome. (b) In the dermoscopic view, we can find a pattern composed of a multifocal delicate pigment network and unfocused arborizing telangiectasias (DermLite Foto; 3Gen, LLC. Original magnification ×10). (c) The result of histologic examination was a piloleiomyoma (haematoxylin and eosin, original magnification ×20).

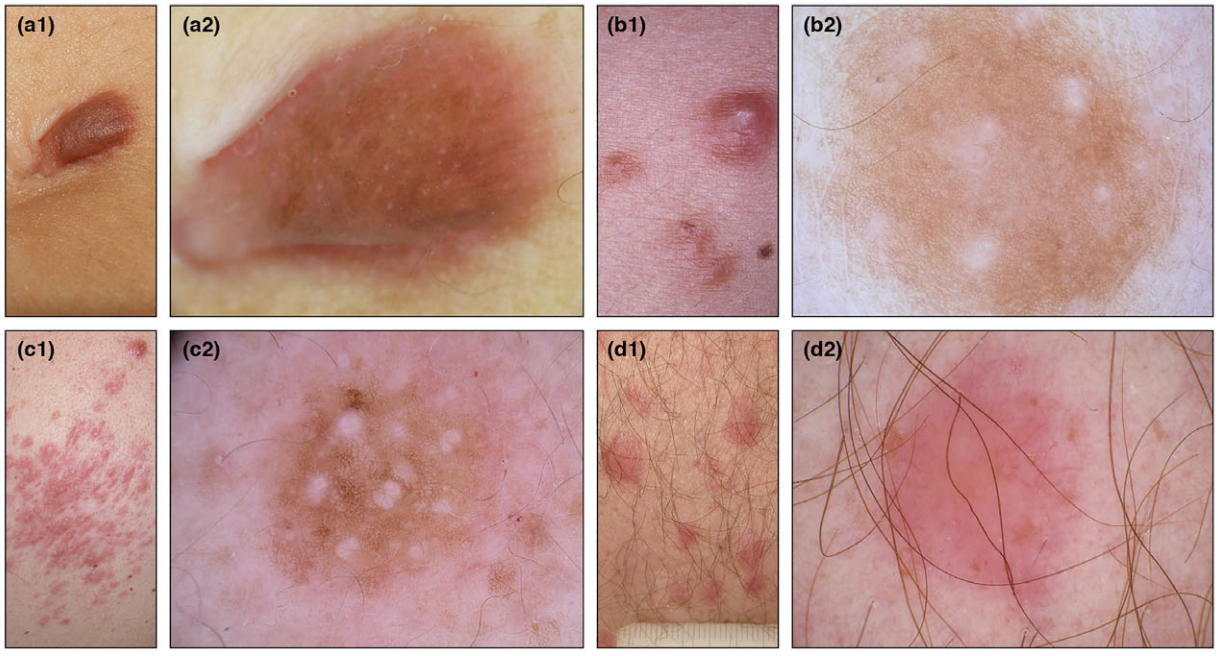

Figure 2 (a1) Piloleiomyoma located on the abdomen of a 42-year-old woman. (a2) In the dermoscopic view, we can find a total delicate pigment network. (b1) Piloleiomyoma located on the back of a 37-year-old man. (b2) In the dermoscopic view, we can find a pattern composed of a total delicate pigment network with multiple hypopigmented areas. (c1) Piloleiomyoma located on the back of a 35-year-old man. (c2) Dermoscopically, we can see the pattern composed of a total delicate pigment network with multiple whitish areas. (d1) Piloleiomyoma located on a leg of a 44-year-old man. (d2) Dermoscopically, we can see a total pinkish homogeneous area with unfocused arborizing telangiectasias which occupies the entire lesion. (DermLite Foto; 3Gen, LLC. Original magnification ×10).

specific. The most common and characteristic pattern associated with AL was the one composed of symmetric pink-reddish tumour with vessels, white structures and the absence of ulceration, which was found in 46.2% of AL (Figs 3, 4), but also in 3.5% of PL, and more importantly in 22.2% of LS. Finally, the most common pattern associated with LS was the one composed of an asymmetric, multilobulated tumour with linear-irregular or polymorphic-atypical vessels and white structures, which was found in 44.4% of cases (Fig. 6), but also in 0.9% of PL and in 15.4% of AL.

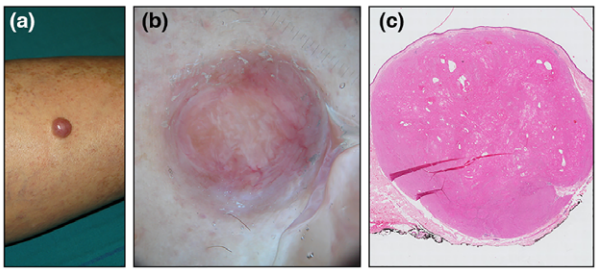

Figure 3 (a) Asymptomatic, reddish papule located on the leg of a 65-year-old woman. (b) In the dermoscopic view, we can find a symmetric lesion with a pattern composed of central white structures and peripheral polymorphous-atypical vessels. The clinical differential diagnosis included cutaneous lymphoma and aneurysmatic dermatofibroma. (DermLite Foto; 3Gen, LLC. Original magnification ×10). (c) The result of histologic examination was a angioleiomyoma (haematoxylin and eosin, original magnification ×20).

Discussion

The results of our study reveal that the pattern composed of a symmetric, total delicate pigment network with the variable presence of multiple hypopigmented areas is the most common dermoscopic pattern associated with PL. This pattern could be identified in 69.3% of our cases of PL (79 out of the 114 lesions) and was not observed either in AL or in LS. The delicate pigment network is defined as the presence of thin lines of light brown colour and regular meshes, regularly distributed and gradually faded into the surrounding skin. This structure was found in 96.5% of our PL and was also the most common feature associated with PL in the literature.[4–8] The histopathological correlation of the delicate pigment network was the presence of a reactive epidermal basal hyperpigmentation; the multiple hypopigmented areas, observed in 49.1% of PL, could correspond histologically to follicular openings or lack of epidermal hyperpigmentation, as we could see in some of our cases. Whitish structures, which should be lighter than normal surrounding skin, were found in 7% of PL and their histopathologic correlation could be the presence of increased or altered collagen in the superficial dermis due to the presence of proliferating smooth muscle cells intermingled with various amounts of collagen bundles, as observed in most of our cases. Vascular structures, mainly unfocused arborizing telangiectasias, were observed in 15.8% of PL and were not representative for most PL. Other features, such as irregular crypts, fingerprint-like structures or a cerebriform pattern, found by Behera *et al.*,[8] were absent in our cases. The clinical differential diagnosis of PL includes angiolipomas, glomus tumour, eccrine spiradenomas and other adnexal tumours, lipomas, neurilemmomas, neurofibromas, dermal nevi and dermatofibromas.[1,2] The only two lesions which may display

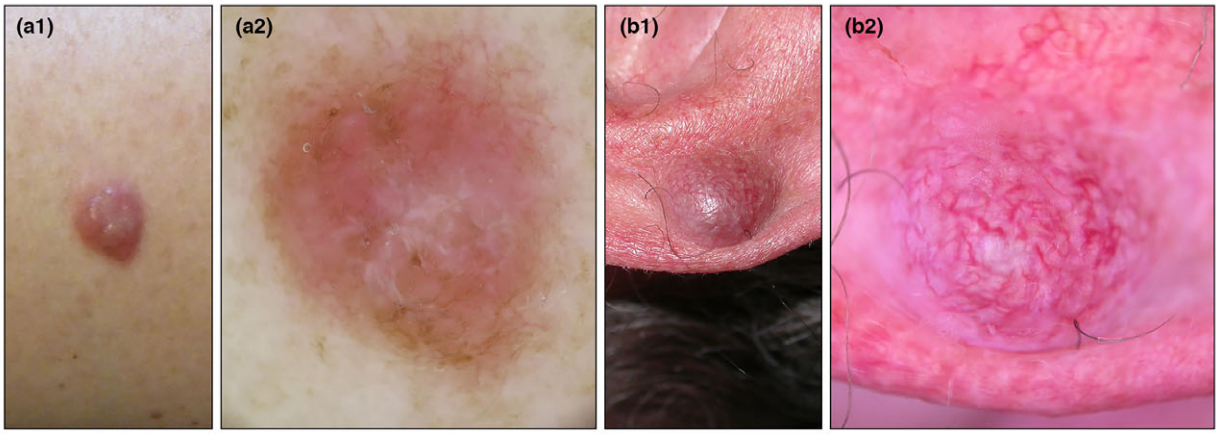

Figure 4 (a1) Angioleiomyoma located on the right arm of a 65-year-old woman. (a2) In the dermoscopic view, we can find the pattern composed of central white structures and milky-red areas and a peripheral delicate pigment network with linear-irregular vessels. (b1) Angioleiomyoma located on the right ear of a 60-year-old man. (b2) Dermoscopically, we can see unfocused arborizing telangiectasias on the whole lesion and central white structures. (DermLite Foto; 3Gen, LLC. Original magnification ×10).

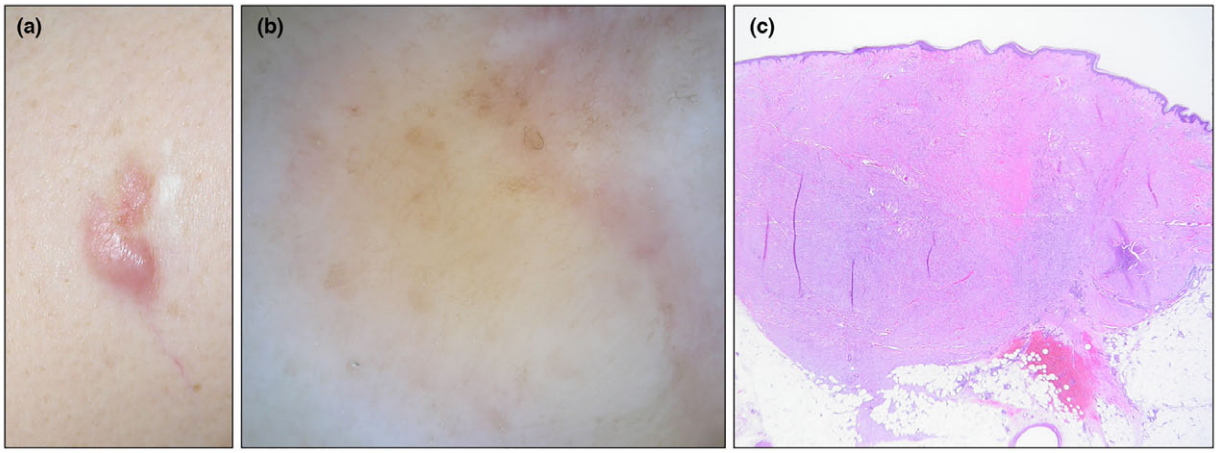

Figure 5 (a) Asymptomatic, multinodular tumour located on the leg of a 65-year-old woman. (b) In the dermoscopic view, we can find an asymmetric tumour with a pattern composed of a multifocal delicate pigment network, whitish structures and dotted vessels. The clinical and dermoscopic diagnosis was dermatofibrosarcoma protuberans (DermLite Foto; 3Gen, LLC. Original magnification ×10). (c) The result of histologic examination was a leiomyosarcoma (haematoxylin and eosin, original magnification ×20).

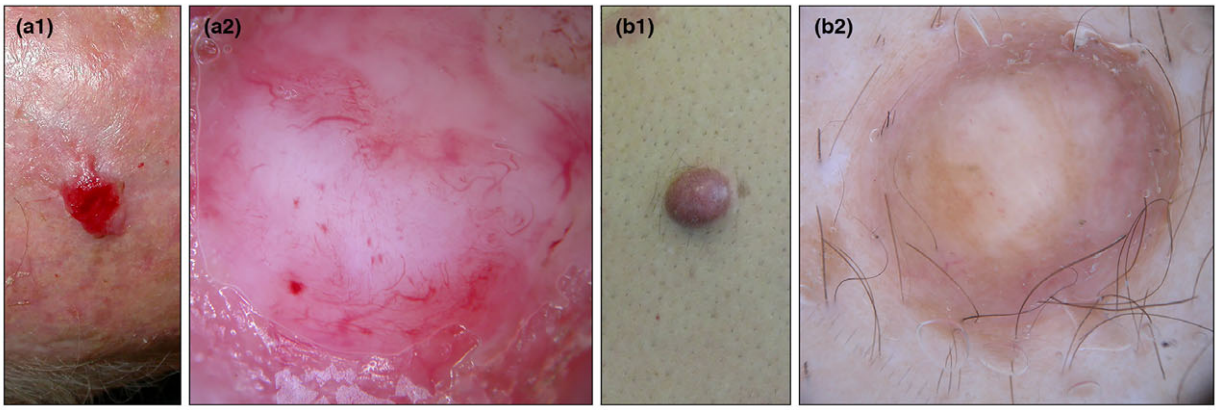

Figure 6 (a1) Leiomyosarcoma located on the scalp of a 91-year-old man. (a2) In the dermoscopic view, we can find polymorphous-atypical vessels in a reddish background with areas of ulceration. (b1) Leiomyosarcoma located on the right leg of a 49-year-old man. (b2) Dermoscopically, we can see the pattern composed of central white structures and a peripheral delicate pigment network with polymorphous-atypical vessels (DermLite Foto; 3Gen, LLC. Original magnification ×10).

dermoscopically a delicate pigment network are neurofibromas, dermatofibromas and mastocytomas.[9–11] Pigmented neurofibromas are usually painless and show dermoscopically the pattern composed of a peripheral delicate pigment network and a central pinkish or whitish structureless area.[10] In our cases, PL were usually painful (78.1%) and showed a delicate pigment network located on the whole lesion. However, neurofibromas and PL can share some dermoscopic patterns. Considering dermatofibromas, Zaballos *et al.*[9] found that 14.6% of cases can show a total delicate pigment network and 34.7% of dermatofibromas display the characteristic pattern composed of peripheral delicate pigment network and central white scar-like patch. Therefore, as referred in the literature, PL can show dermoscopic features overlapping with dermatofibromas. However, it is important to note that the great majority of dermatofibromas are painless and most PL are painful.

The dermoscopic features associated with AL were more variable and less specific. The most common pattern associated with AL was the one composed of symmetric pink-reddish tumour with vascular and white structures, which was found in 46.2% of AL, but also in 3.5% of PL, and more importantly in 22.2% of LS. The histopathologic correlation of the total reddish structureless

area and vascular structures is the presence of the interlacing bundles of uniform smooth muscle cells, distributed around numerous vessels with walls of variable thickness, characteristic of AL. White structures may correspond to the changes in the orientation of collagen due to the pressure of the large dermal tumour. Delicate pigment network, a very common feature of PL, was found in 30.8% of cases and was always distributed peripherically. However, it is important to emphasize that 23% of AL were asymmetric, 15% presented as multilobulated tumours, 66% of the vascular structures were linear-irregular vessels or polymorphic-atypical vessels and were presented multifocally, and milky-red areas were detected in 61% of cases. As a result of its dermoscopic variability and alarming features, AL are usually difficult to recognize and easy to misdiagnose as other benign or malignant lesions, even melanoma.[12–14] In our study, none of the AL was correctly diagnosed and, more importantly, 61% of the cases were dermoscopically diagnosed as a malignant tumour.

The dermoscopic structures associated with LS were even more variable and less specific than those associated with AL. However, most of the dermoscopic features of LS were highly suggestive of malignancy (77.8% were asymmetric, 55.6% were ulcerated, 55.6% were multilobulated, the pigment network was atypical in 40% of cases and multifocal in 80% of cases, the majority of vascular structures (62.5%) were polymorphic-atypical vessels, 55.6% of cases showed milky-red areas, and 88.9% of cases display white structures, including chrysalis in 37.5%). The most common pattern associated with LS was the one composed of an asymmetric, multilobulated tumour with linear-irregular or polymorphic-atypical vessels and white structures, which was found in 44.4% of cases. These dermoscopic features are associated with malignant tumours such as melanoma,[14] Merkel cell carcinoma,[15] angiosarcoma,[16] dermatofibrosarcoma protuberans,[17] porocarcinoma[12] and B-cell lymphomas[18,19] to cite only a few. In fact, in our study, none of the LS was correctly diagnosed but all but one case were diagnosed as malignant.

Our study has several limitations. First, it was conducted retrospectively. Second, the histopathologic diagnosis was based on the official pathology diagnosis from the corresponding centre; a second pathologist did not confirm the diagnoses. Third, our study was not designed to analyse the sensitivity and specificity of each dermoscopic structure or pattern for the diagnosis of smooth muscle tumours. Finally, we have to warn the reader that the presence of some signs depends on the technique used (polarized vs. non-polarized); white structures (including chrysalis) are not seen on non-polarized dermoscopy, and the visualization of vascular structures is influenced by the pressure applied on the skin with the dermatoscope.

In conclusion, the pattern composed of a symmetric, total delicate pigment network with the variable presence of multiple hypopigmented areas in a painful lesion is the most common and characteristic dermoscopic pattern associated with PL. The dermoscopic patterns associated with AL and LS are more variable and less specific, and many tumours can show them. However, dermoscopy helps to diagnose the latter as a malignant lesion. Therefore, dermoscopy can be considered useful as an adjuvant diagnostic tool that provides some extra information about cutaneous smooth muscle neoplasms. However, we are in favour of removing not only all LS but also all AL to study them histopathologically because this tumour is also a simulator of other more serious lesions, including melanomas, on clinical and dermoscopic examination.

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