

Cutaneous Leiomyoma: Novel Histologic Findings for Classification and Diagnosis

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Abstract- Smooth muscle tumors rather benign or malignant can arise wherever the muscular tissue presents but cutaneous leiomyoma is one of the rare benign tumors of the which even the diagnostic criteria from the malignant type of the tumor is still in doubt. This study was aimed to compare the subtypes of cutaneous leiomyoma from different histologic aspects in order to find unique criteria for better classification and diagnosis. The six year data base of our center was reviewed and 25 patients with cutaneous leiomyoma were included in this study. Of 25 patients, 5 were female and 20 were male. 5 patients had angioleiomyoma (ALM) and 20 had pilar leiomyoma (PLM). ALM had following characteristics: dilated vascular canals intermingled with compact smooth muscle bundles; well circumscribe counter and myxoid and hyaline changes through the tumor. In contrast, PLMs had following histologic features: poor defined outline, entrapped hair follicles and eccrine glands, acanthosis and elongated rete ridges with hyperpigmentation and smooth muscle bundles which are interdigitated with elongated rete ridges. Here we introduced some distinct histological features for each subtype of the cutaneous leiomyoma which can lead to create novel criteria for classification and diagnosis of the lesion.

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Introduction

Smooth muscle tumors both benign and malignant can arise wherever the muscular tissue presents which uterine is a very common place for the tumor (1,2). Cutaneous leiomyoma is one of the rare benign tumors of the skin and the diagnostic criteria for distinguishing benign tumors from malignant types are still in doubt (3). Sometimes immunohistochemical (IHC) studies are used in order to confirm the diagnosis of the cutaneous leiomyoma (4). Patients follow up is at last the most reliable factor that can prove the benign or malignant theme of the tumor. The cutaneous leiomyoma can be subclassified as pillar leiomyoma (PLM) which arises from erector pili muscle and angioleiomyoma (ALM) which arises from muscularis layer of vessels in different organs and finally genital leiomyoma (GLM) (1,5).

In this study we introduced 25 cases of cutaneous leiomyoma and discussed about some criteria for better histologic classification of the tumor.

Patients and Methods

Study

We reviewed our database in Razi dermatology hospital, Tehran, Iran, and 25 patients with cutaneous leiomyoma were enrolled in this study. The data were obtained from our medical records during January 2008 to February 2011 and all patients' pathologic samples were again examined in order to confirm the diagnosis. All the patients were healthy and no clinical complication had occurred. None of the patients had family history of cutaneous leiomyoma and also none of the patients with multiple cutaneous leiomyomas had extracutaneous leiomyoma. IHC study for desmin and smooth muscle antigen (SMA) was also performed for diagnosis confirmation.

Results

Clinical characteristics

The main characteristics of the patients are summarized

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in Table 1. 80% of the patients were male (20 male *versus* 5 female). Mean age of diagnosis was 44.6 ± 13.6 in male and 50.8 ± 14.6 in females, with no statistically significant difference. The most common place was extremities (52%). The sites of tumors are shown in Table 1. The period of having the lesions before diagnosis ranged from 2 months up to 20 years. Of 25 specimens, 19 were PLM, one was GLM and 5 were ALM. There was no significant difference between tumor subtypes in gender, age, site of occurrence, multiplicity, size of the lesion and painfulness. The tumor size was ranged from 2.8 mm up to 10.5 mm with average of 5.5mm as measured with micrometer. Among 25 patients, 4 (16%) had multiple lesions which one of them was female and three were male (Figure 1). All the five female patients (100%) were presented by pain in comparison to 8 out of 20 male patients (40%), which the difference was statistically significant ($P=0.03$), totally in 13 patients (54.1%) the only symptom was pain. Indeed there was statistically significant difference

between single *versus* multiple lesions which all multiple lesions were painful ($P=0.04$). There was no significant difference between male and female patients in location of the lesion, size or multiplicity.



Figure 1. A patient with multiple leiomyoma.

Table 1. Clinical features of 25 patients diagnosed as leiomyoma.

Numerical order	Sex	Age	Location*	Duration (M,Y)	Solitary/ Multiple	Pain	Diagnosis
1	M	65	LL	2 Y	S	-	PLM
2	M	53	A	5 Y	S	-	PLM
3	F	60	LL	6 M	S	+	PLM
4	M	35	S	1 Y	S	-	PLM
5	M	66	F	NA	S	-	PLM
6	M	30	LL	15 Y	S	+	PLM
7	M	52	UL	7 M	S	-	PLM
8	M	50	F	NA	S	-	PLM
9	M	52	N	20 Y	S	-	PLM
10	M	38	G	10 M	S	-	GLM
11	M	45	F	2 M	S	+	PLM
12	M	27	F	2 Y	S	+	PLM
13	M	75	LL	NA	S	-	PLM
14	M	46	LL	NA	S	-	PLM
15	F	25	UL	1 Y	S	+	PLM
16	M	47	N	6 Y	S	-	PLM
17	F	59	UL	10 Y	M	+	PLM
18	M	45	T, UL	9 Y	M	+	PLM
19	M	31	T	NA	M	+	PLM
20	M	44	F, UL, LL	18 Y	M	+	PLM
21	F	56	T	NA	S	+	ALM
22	M	34	LL	18 Y	S	+	ALM
23	F	54	UL	9 M	S	+	ALM
24	M	34	LL	9 Y	S	+	ALM
25	M	24	F	4 Y	S	-	ALM

* A, Axilla; G, Genitalia; F, Face; LL; Lower limb; N, Neck; S, Scalp; T, Trunk; UL, Upper limb.

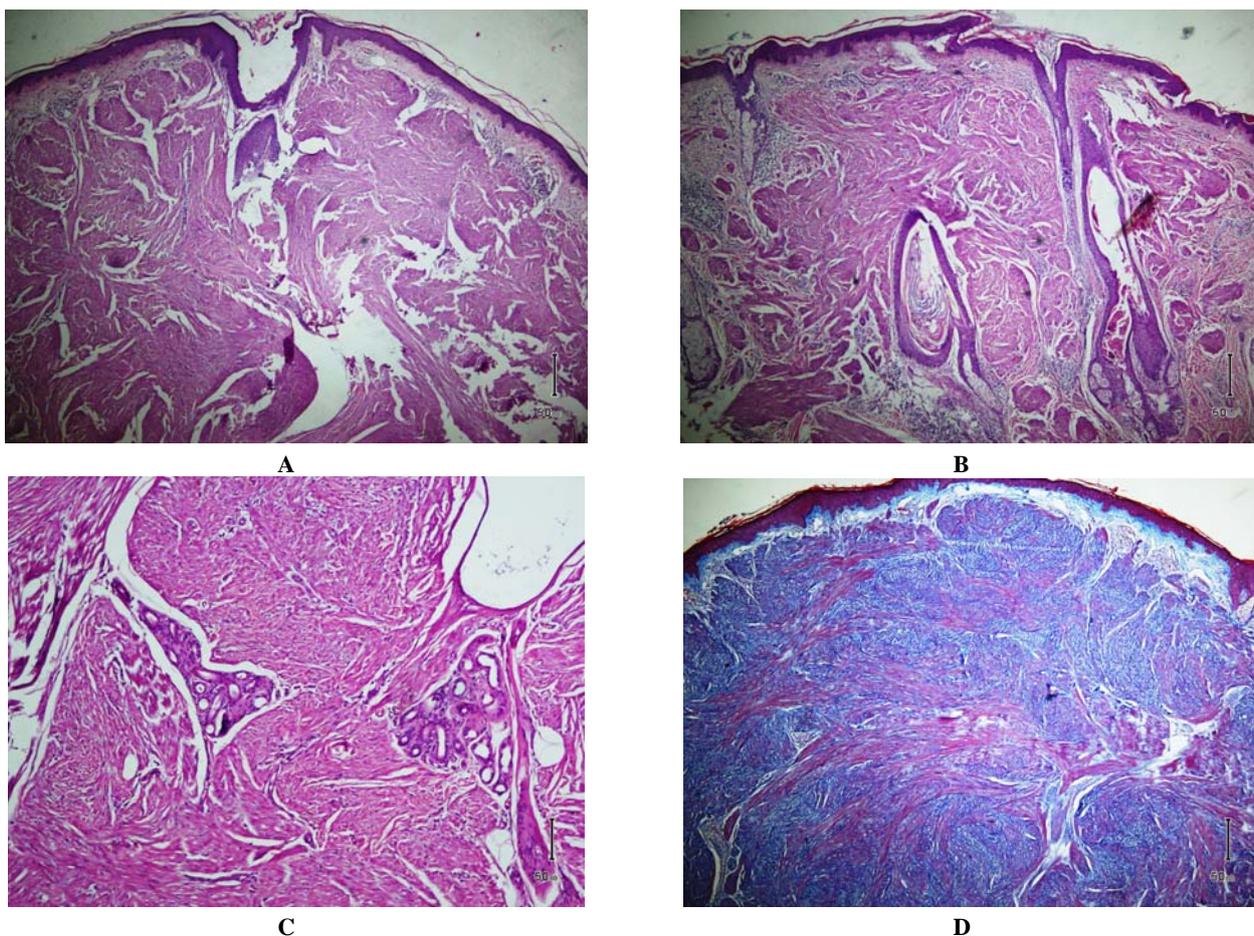


Figure 2. A. PLM in upper dermis, B. High magnitude of the same tumor, C. Showing eccrine gland entrapping in PLM, D. Trichrome staining showing muscle bundles (muscle bundles are in red stain).

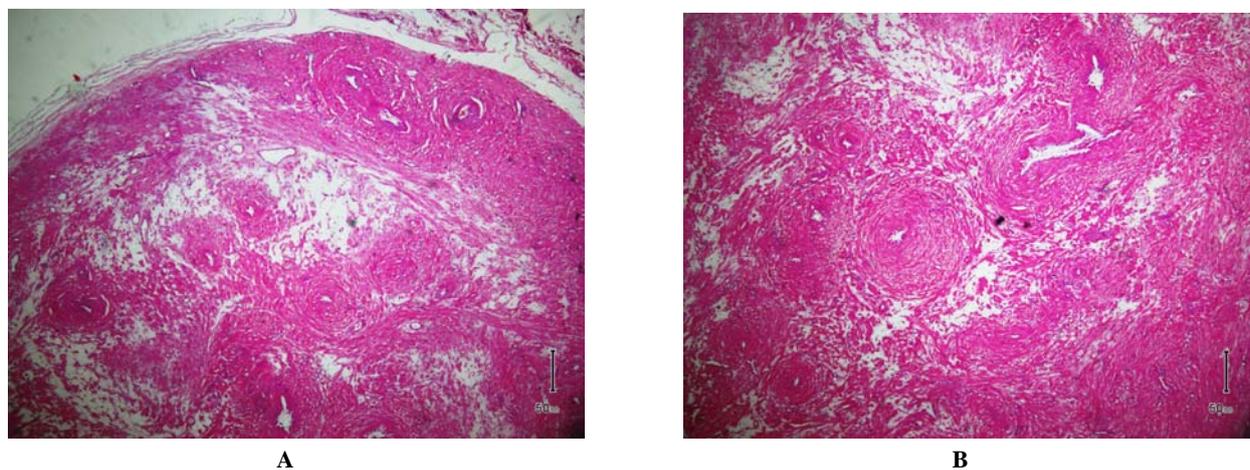


Figure 3. A. ALM with well defined smooth contour, B. Showing thick walled vessels and myxoid changes.

Histopathology examination

Pathological examination of all the specimens revealed interlacing short fascicles of bland spindle cells with elongated nuclei and blunt ends. Two subtypes of cutaneous leiomyoma were diagnosed, PLM and ALM. As histologic features of GLM are completely similar to

PLM, it is categorized as PLM in our analysis.

PLMs were placed mostly in upper dermis extending to deep dermis but all ALMs were placed in deep dermis (Figure 2A-D). All ALMs were well circumscribed but all PLMs revealed ill defined outlines with smooth muscle extension through the main tumor.

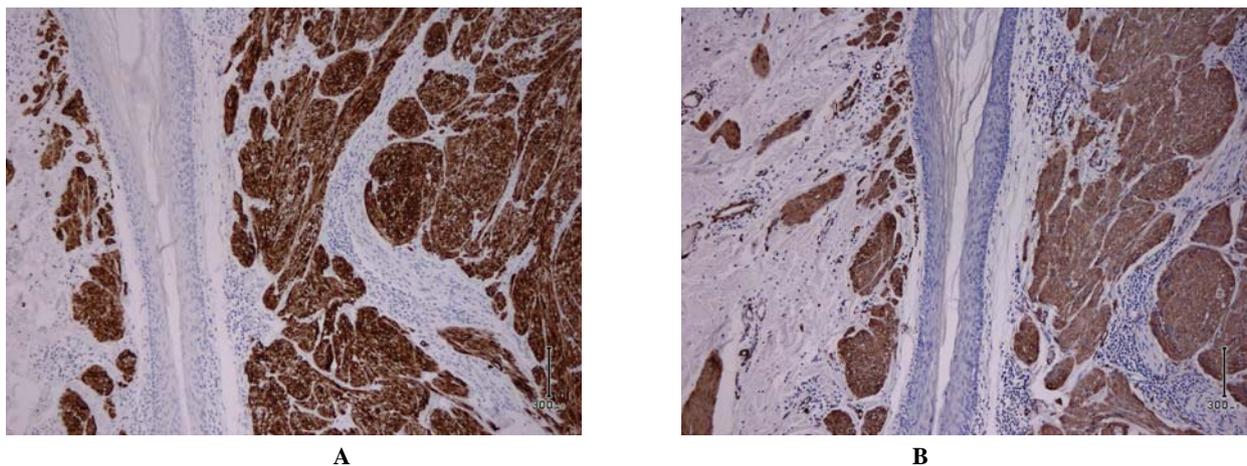


Figure 4. A, B. IHC staining for desmin and SMA.

Table 2. Pathologic characteristics of 25 LM.

Pathologic Diagnosis (number)	Diameter (mean)/mm	Entrapped hair follicle	Entrapped eccrine Gland	Acanthosis	Pigmented rete ridges	Hyaline/myxoid change
Solitary PLM (n=16)	6.9	9/16	10/16	13/16	12/16	0
Multiple PLM (n=4)	5	3/4	2/4	2/4	3/4	0
ALM (n=5)	5	0	0	0	0	3/4

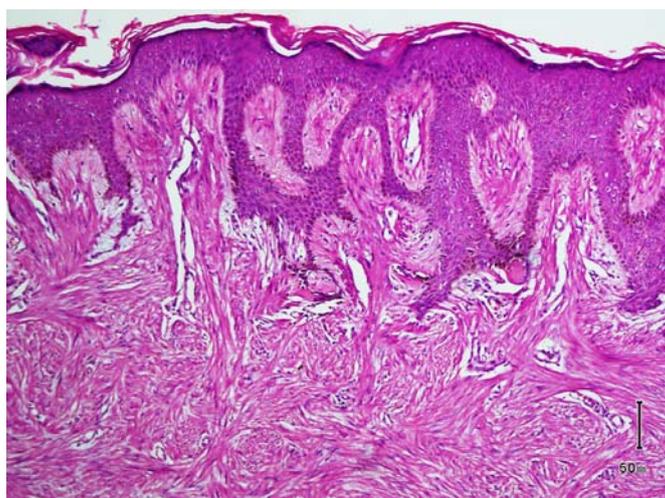


Figure 5. Showing muscle bundles interdigitating with rete ridges.

In ALM, there was slit or dilated vascular channels intermingled with closely compact smooth muscle bundles (Figure 3A and B). Histologic evaluation among five ALMs revealed two solid, two cavernous and one venous type as described in previous articles (6). All the specimens showed positive reactivity for desmin and SMA, IHC markers used to confirm the diagnosis (Figure 4A and B).

The above mentioned histologic features are mentioned before and in other literature but here we

introduced some novel histologic features for better distinguishing ALM from PLM which are not mentioned elsewhere. Entrapped hair follicles and eccrine glands were seen in most PLMs (>50%) but never in ALMs; 75% of ALMs revealed myxoid and hyaline changes through the tumor, whereas none of PLMs presented these changes. Acanthosis and elongated rete ridges with hyperpigmentation were seen in 75% of PLM but not in ALM (Table 2). Smooth muscle bundles which are interdigitated with elongated rete ridges were also a

histological feature purely seen in PLM but not in ALM (Figure 5).

Discussion

Leiomyomas are benign tumors but very rare in skin (7). They arise mostly from pilar muscles but can also arise from any tissue containing muscular tissue *e.g.* blood vessels, areola or external genitalia, which is the logic of denomination of the lesions as ALM, PLM and GLM (5). Clinically they appear as reddish to brownish papules and are less than 15 mm in diameter (8). Most of the tumors arise in limbs except thigh, which according to literature is a rare place for tumor occurrence. Our patients' clinical presentations are in accordance with literature, which more than fifty percent of the patients had lesions on their extremities (9). Most of the reported cases are male and our results are in line with the mentioned reports. According to our results about eighty percent of the patients presented with a single lesion which is in line with previous reports (10).

Histologically, leiomyomas are well differentiated and mostly encapsulated tumors with scant nuclear atypia or mitosis in contrast to malignant variant of the tumor, leiomyosarcoma, which presents with high nuclear atypia and mitotic activity rate (10). According to our cases the leiomyomas can present with mild mitotic activity which is mentioned in some other reports, but nuclear pleomorphism was not mentioned.

Here we introduced some histological features in both types of the cutaneous leiomyomas which were not previously mentioned elsewhere; Pigmented rete ridges, acanthosis, entrapped eccrine glands and hair follicles were findings purely seen in PLM but not in ALM, in contrast hyaline/myxoid changes within tumor was seen in ALM but not in PLM. Another histological feature which is introduced here is interdigitated smooth muscle bundles with elongated rete ridges which are noted in PLM but not in ALM.

Cutaneous leiomyomas are usually asymptomatic but can cause pain due to nerve bundles compression or contraction of tumoral muscle bundles within the skin (11). About half of our patients presented with pain which is noted up to 90 percent in different papers (8,12). Our results also showed that female patients (gender) and multiplicity of the lesion may be associated with the painfulness of the lesion.

The patients underwent surgery and none of our cases had recurrence. According to painful presentation of the lesion, the cutaneous leiomyoma should be come in to differential diagnosis of painful nodules of the skin

(2). Lipoma, ganglion, fibromatosis (desmoids tumor), schwannoma, haemangioma, foreign body granuloma, pseudoaneurysm, inclusion cyst, giant cell tumour of tendon sheath and glomus tumor are some differential diagnosis of this cutaneous nodular lesion (13). It should be mentioned that in most of the cases the leiomyoma is not one of the preoperative differential diagnosis of the nodule (only 6 out of 25 cases were clinically diagnosed as cutaneous leiomyoma in this study) which shows rare possibility of preoperative diagnosis and the importance of increasing awareness about the lesion, taking biopsies from cutaneous lesions and more usage of imaging in preoperative examination. (6,13).

Totally, to our knowledge, the mentioned histological findings are not mentioned elsewhere and we introduced some histological features in both types of the cutaneous leiomyomas which were not previously mentioned elsewhere. Although there is no difference in treatment of the both subtypes but presenting characteristic histologic features and definite diagnosis is of scientific and academic value.

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