ISSN 2473-4799

DERMATOLOGY
Open Journal

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Brief Research Report

Lichen Planus Pigmentosus in North Africa: A Series of 17 Cases

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Article information
Received: March 18th, 2019; **Revised:** April 15th, 2019; **Accepted:** May 7th, 2019; **Published:** May 15th, 2019

Cite this article
Benchikhi H. Lichen planus pigmentosus in north Africa: A series of 17 cases. *Dermatol Open J.* 2019; 4(1): 10-14. doi: 10.17140/DRMTOJ-4-135

ABSTRACT

Background
Lichen planus pigmentosus (LPPig) is considered as a rare macular and pigmented variant of lichen planus. It is mainly reported in Indian and Latino American patients.

Objective
The aim of this paper is to report the characteristics of this condition in Moroccan patients.

Patients and Methods
Patients attending a private dermatology practice, in Casablanca, Morocco, and presenting with LPPig were consecutively enrolled. Inclusion criteria were the presence of macular pigmented lesions of the face, neck or arms, and/or cutaneous histology showing pigment incontinence with basal cell vacuolization and lichenoid infiltrate. Patients with melasma or post-inflammatory pigmentation were excluded. Demographic and medical data were collected.

Results
From January 2015 to December 2018, 17 patients were included, one man and 16 women, mean age 57.4±11 years. Ten patients had phototype III and five phototype IV. The mean duration of the disease was 6.7±7 years. The lesions were located on the face in all patients, with the involvement of the perioral area in 13 cases (76.4%), the forehead in 9 cases. The neck was involved in 9 cases (53%). In 7 cases (41%), frontal fibrosing alopecia was observed. Three patients had thyroiditis, three others had diabetes mellitus. Histology was performed in six cases, showing pigment incontinence and basal vacuolization. Lichenoid infiltrate was observed in four cases. Eleven patients were given tranexamic acid per os, 500 mg twice daily for 3 or 6 months. Topically, all patients but one received hydroquinone, and sixteen high potency dermocorticoids to the lesions. An the follow-up, only four patients had an excellent result with complete resolution of the lesions, in the others, the lesions stayed similar.

Conclusion
LPPig exits in North Africans although its prevalence is probably underestimated. This pigmentary disorder remains a challenging disease for therapy.

Keywords
Lichen planus pigmentosus; Tranexamic acid.

INTRODUCTION

Lichen planus pigmentosus (LPPig) is considered as a rare macular and pigmented variant of lichen planus.[1] Two other conditions are similar, with close clinical and histological characteristics: Ashy dermatosis described by Ramirez et al in 1957[2] and "pigmented cosmetic melanosis" also called Riehl's melanosis by Nakayama.[3] This pigmented dermatosis is characterized by brown to grey macules, located in face, neck, arms, and more rarely in flexural areas. Histology shows pigment incontinence and, sometimes, basal cell vacuolization. The disease affects preferentially women, in young to middle ages and predominantly with dark skin phototypes. LPPig was first described in Latin America, Indian subcontinent, the middle east, and the far east but never reported in North Africa. The aim of this paper is to report the characteristics of this condition in Moroccan patients.

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PATIENTS AND METHODS

The study was performed on patients attending the private practice of dermatology, in Casablanca, Morocco, from January 2015 to December 2018. Inclusion criteria were the presence of macular pigmented lesions of the face, neck or arms, and/or cutaneous histology showing pigment incontinence with basal cell vacuolization and lichenoid infiltrate. Patients with melasma, post-inflammatory pigmentation or photosensitization were excluded. Following data were collected: Age, gender, duration of disease, location, histology, associated diseases, treatment or physical procedures, and follow-up.

RESULTS

Seventeen patients were enrolled, one man et 16 women, mean age 57.4±11-years, from 31 to 80-years. Ten patients had phototype III, five phototype IV and two phototypes V. The mean duration of the disease was 6.7±7 years, from one to 25-years (Table 1).

Table 1. *Demographic and Clinical Data of 17 Patients with Lichen Planus Pigmentosus*

N°	Sex	Age (years)	Phototype	Duration (years)	Face	Peribucal	Neck	FFA	Histology	Tranexamic Acid	Follow up
1	M	31	III	4	+	+	+	-	+	+	X
2	F	80	V	2	+	+	-	-	-	-	Unfavorable
3	F	63	III	7	+	+	-	-	+	-	Good
4	F	50	III	3	+	+	-	-	+	-	X
5	F	52	IV	6	+	+	-	-	-	-	X
6	F	68	V	25	+	+	+	+	+	-	Unfavorable
7	F	43	III	2	+	+	+	-	-	+	X
8	F	53	III	3	+	+	-	-	-	-	X
9	F	52	III	12	+	-	-	-	-	+	X
10	F	45	III	4	+	-	+	+	-	+	Good
11	F	59	III	20	+	-	+	+	-	+	Good
12	F	67	III	1	+	-	+	+	-	+	Good
13	F	68	IV	1	+	+	+	+	-	+	Still being treated
14	F	43	IV	3	+	+	-	+	-	+	Still being treated
15	F	55	IV	20	+	+	-	-	-	+	Still being treated
16	F	55	III	1	+	+	+	+	+	+	Unchanged
17	F	42	III	0,5	+	+	+	-	+	+	Still being treated

FFA: Frontal Fibrosing alopecia, X: Unknown

The face was the initial location in all patients, followed by the neck. At the examination, the lesions were located in the face in all patients, with the involvement of the peribuccal area in 13 cases (76.4%), the forehead in 9 cases (53%) and the temples in 2 cases (Figures 1 and 2). One patient had also bilateral hyperpigmentation of eyelids and one had cheilitis. The neck was involved in 9 cases (53%), the dorsum of the hands in one case; no flexural lesion was observed. The color of the lesions was brown or grew. No inflammatory lesion was found 7 cases (41%), lichen planopilaris was associated, type of frontal fibrosing alopecia with the involvement of the eyebrows in three cases (Figure 3). Three patients had thyroiditis with hypothyroidism in one case; three other patients had diabetes mellitus. All patients had intense sun exposure but no fragrance application was reported.

Six patients underwent cutaneous histology; pigment incontinence was observed in all cases, as basal vacuolization (Figure 4). The biopsy showed lichenoid infiltrate.

Eight patients received antimalarials before 2016; after this date, 11 patients had tranexamic acid orally, 500 mg twice daily during 3 or 6-months. One patient had Vibramycin during 3-months and another one had an injection of corticosteroids. Typically, all except one patient received hydroquinone and sixteen high potency corticosteroids.

Figure 1. *Lichen Planus Pigmentosus. Diffuse Brown Macules of the Face*

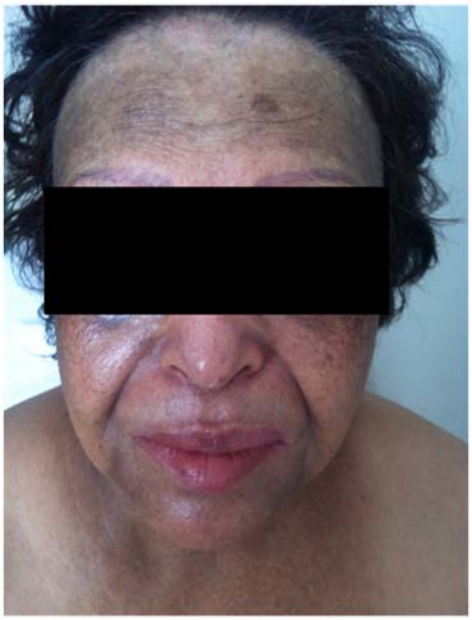

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Figure 2. *Lichen Planus Pigmentosus. Grey Patchy Pigmentation of the Submental Area*

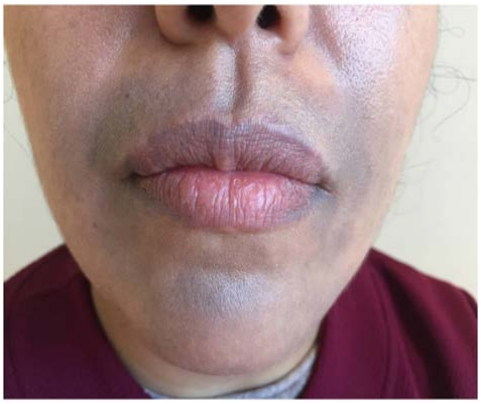


Figure 3. *Lichen Planus Pigmentosus and Frontal Fibrosing Alopecia*

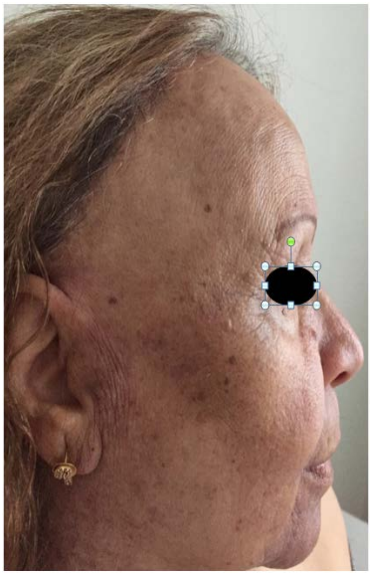
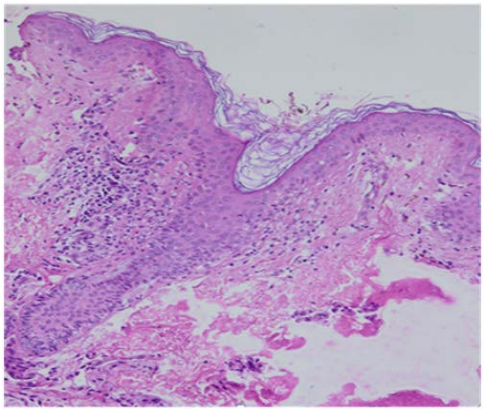


Figure 4. *Lichen Planus Pigmentosus, Cutaneous Histology: Basal Vacuolization, Focal Lichenoid Infiltrate, Pigmentary Incontinence. Hematoxylin And Eosine X100*



On the follow-up, four patients had an excellent result with complete resolution of the lesions, three of them received tranexamic acid during a course of three months. Five other patients still received treatment; two had less favorable evolution and in the six remaining patients, lesions stayed similar.

DISCUSSION

We report herein a series of 17 Moroccan patients with LPPig. Up till now, most cases were described in patients from India, Latin America and Middle East regions and this series are the first one reported in North Africa. This condition is considered as an uncommon variant of lichen planus; it was reported in young to middle-aged adult females of dark skin phototypes. Most of our patients were phototypes III or IV with a clear female predominance. In LPPig, the lesions are irregularly shaped or oval, brown to gray macules or patches, located on sun-exposed areas or, less commonly, on intertriginous folds. In our series, the face was involved in all cases, mainly in the perioral area followed by the neck. Cutaneous lesions were overall bilateral with a patch pattern. We noticed only one case with associated lesions on the hands but no flexural lesions. Bilateral upper eyelid lesions can be, although rarely, the predominant location in LPPig.[4] Other rare clinical forms are reported in the literature as blaschkoid, zosteriform, segmental or mucosal LPPig.[5,6] Most of our patients had a chronic course of their disease and this is a common feature in LLPig. Triggering factors were sun exposure with photoaggravation in some cases,[7] presence of hepatitis C virus and application of mustard oil.[6,7]

LPPig may be associated with other conditions. In our series, seven patients had frontal fibrosing alopecia (FFA). The first case of LPPig in an FFA patient was described by Dlova.[8] Two recent papers reported a strong association between FFA and LPPig. Among 91 FFA patients reported by Mervis, 45% were of Hispanic/Latino origin, and were more likely to have associated LPPig than those of other ethnicity.[9] There was a strong statistical association between women with FFA and LPPig[10,11] and in our series, near fifty percent of LLPig patients had FFA, confirming that the is a spectrum between LLPig and FFA. One case of associated LLPig and nail lichen planus was reported.[12] As in common lichen planus, some autoimmune conditions seem to be associated with LLPig, mainly diabetes mellitus, and thyroid disorders.

Many authors consider LPPig as a similar condition to Ashy dermatosis because of clinical and histological overlapping features.[13] In fact, Ashy dermatosis, also renamed "*erythema dyschromicum perstans*", is characterized by slowly progressive ashy-colored hyperpigmentation on face, arms, neck and trunk. Cases were reported in Latin American and Asian patients with a female predominance.[2] In both conditions, histology shows vacuolization of the basal cell layer, a slight mononuclear infiltrate in the upper dermis with dermal melanophages. Lichenoid reaction is more commonly seen in LPPig (90%) than in Ashy dermatosis (57%).[14] In our series, histology was performed only in six cases and showed identical features. So, the utility of histology is weak to distinguish between both conditions,[15] thus, avoiding a biopsy scar in the face is preferable. On immunohistology, direct immunofluorescence (DIF) staining was positive in 6 out of 21 cases of LPPig by Thientavorn, the most common pattern being immunoglobulin M (IgM)

colloid bodies. In the same series, patch testing was performed and was positive in 40 and 36.36% of Ashy dermatosis and LPPig cases respectively.[14] Although the controversy is still going, the two conditions have more similarities than differences.[13,15,16]

The diagnosis of LPPig is very difficult and can be confused with melasma or post inflammatory hyperpigmentation. Clinical signs are very close and histology is not systematically performed on the face. In our country, melasma remains the most common and the most challenging hyperpigmentation of the face.[17] Clinically, it seems that melasma predominates on the cheeks but spares the perimental area while it is the contrary in LLPig. In our series, the most frequently involved area is the peribuccal one then the neck, which is not so common in melasma. Thus, using other procedures to differentiate between the two conditions is useful. Dermoscopy can show discrete bluish-grey dots, globules, blotches and rods against a brownish background, these are typical features in LPPig according to Gajjar.[5]

Treatment of acquired hyperpigmentation has limited success, especially in LPPig. Although four patients had a total clearance of the patches, the remaining ones still have slight or no improvement. Topical treatments may involve high potency corticosteroids, hydroquinone and tacrolimus. Tacrolimus ointment 0.03% showed a good lightening of the disease after a course of 12-weeks in the series of Al Mutairi.[18] Among systemic treatment, hydroxychloroqine was commonly used with slight improvement. As in other pigmentary disorders, tranexamic acid is recently used in this condition but result has to be confirmed in LPPig. However, in our series, tranexamic acid seems to be a promising treatment.

CONCLUSION

LPPig remains a challenging disease, not only for clinical diagnosis, but also for therapy. In North Africa, especially in Morocco, the prevalence of this condition is probably underestimated.

ACKNOWLEDMENT

Ackowlegments to Pr Faridi Marnissi et Pr Abdelmajid Rifki for microphotos.

CONSENT

The authors have received oral informed consent from the patient whose photographs are included in the manuscript.

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