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Case Report

Exogenous Ochronosis in Facial Lichen Planus Pigmentosus

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Received Date: 06-03-2021; Accepted Date: 23-04-2021; Published Date: 30-04-2021

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Abstract

Lichen planus pigmentosus is a chronic inflammatory pigmentary dermatosis considered a rare variant of the cutaneous lichen planus. It is characterized by acquired dark brown to gray macular pigmentation involving sun-exposed areas commonly found in dark-skinned patients. Exogenous ochronosis is an acquired cause of blue-gray to black pigmentation involving the skin and mucous tissues. We report a case of exogenous ochronosis occurring in a 45-year-old patient treated for lichen planus pigmentosus. She was treated with Kligman formula containing hydroquinone. The patient presented to our Department for progressive worsening of the malar pigmentation. Dermoscopy revealed gray to black rounded and homogeneous areas with arciform structures obliterating the follicular openings and telangiectatic vessels making us suspecting exogenous ochronosis confirmed by histology.

Keywords

Lichen Planus Pigmentosus; Exogenous Ochronosis; Dermoscopy; Histology

Kadiri SE | Volume 2; Issue 1 (2021) | JDR-2(1)-021 | Case Report

Citation: Kadiri SE, et al. Exogenous Ochronosis in Facial Lichen Planus Pigmentosus. J Dermatol Res. 2021;2(1):1-7.

DOI: <http://dx.doi.org/10.46889/JDR.2021.2106>

Introduction

Lichen planus pigmentosus is a chronic inflammatory pigmentary dermatosis considered a rare variant of the cutaneous lichen planus. It is characterized by acquired dark brown to gray macular pigmentation involving sun-exposed areas commonly found in dark-skinned patients. Exogenous ochronosis is an acquired cause of blue-gray to black pigmentation involving the skin and mucous tissues. We report a case of exogenous ochronosis occurring in a patient treated for lichen planus pigmentosus.

Patient and Observation

A 45-year-old postmenopausal, phototype IV, presented with a history of 10 years of frontal recessing line, eyebrow loss, and brown-gray pigmentation of both cheeks. A skin biopsy of both pigmented lesions and scalp alopecia was compatible with the diagnosis of lichen planus pigmentosus and frontal fibrosing alopecia. The patient was treated with chemical photoprotection, doxycycline, and Kligman preparation containing hydroquinone 5%, retinoic acid 0.2%, hydrocortisone 1%, and vitamin C for four months. The follow-up was lost. She applied the treatment by self-medication for two years without interruption. She presented to our Department for progressive worsening of the malar pigmentation. Dermatological examination showed hyperchromic, gray-brown macules in the zygomatic areas with frontal recessing line and eyebrow loss (Fig. 1 and 2). Dermoscopy revealed gray to black rounded and homogeneous areas with arciform structures obliterating the follicular openings and telangiectatic vessels (Fig. 3 and 4) making us suspecting exogenous ochronosis. Dermoscopy showed also regularly distributed brown-grey globules and dots compatible with a facial lichen planus pigmentosus (Fig. 5).

Histopathological examination revealed ochronotic pigment represented by ovoid bodies and bananas in the dermis compatible with the diagnosis of exogenous ochronosis (Fig. 6). We forbade the use of topical depigmenting agents containing hydroquinone.

Figure 1: Hyperchromic, grey-brown macules in the zygomatic areas with frontal recessing line and eyebrow loss.

Figure 2: Hyperchromic, grey-brown macules in the zygomatic areas with frontal recessing line and eyebrow loss.

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Figure 3: Exogenous ochronosis. Dermoscopy showing grey to black rounded with arciform structures (blue triangle).

Figure 4: Exogenous ochronosis. Dermoscopy showing arciform structures and telangiectatic vessels.

Figure 5: Lichen planus pigmentosus. Dermoscopy showing regularly distributed brown-grey globules and dots.

Figure 6: Histology showing ochronotic pigment represented by a banana-like structure (red triangle).

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Discussion

Ochronosis was first described by Virchow in 1866 to describe a blue-black cutaneous pigmentation in the skin associated with an ochre pigment deposition in microscopy [1]. They are two main types: alkaptonuria and exogenous ochronosis. Alkaptonuria is an autosomal recessively inherited condition due to deficiency of Homogentisate 1,2-dioxygenase (HGD) in the liver and kidneys leading to an accumulation of homogentisic acid in the skin, cartilage, and fibrous tissues [2]. Exogenous ochronosis has been reported following the long-term use of bleaching creams containing hydroquinone, resorcinol, picric acid, mercury, and antimalarials is a chronic dermatosis characterized by the presence of blue-black or grey-blue pigmentation of the skin and mucous tissue. This condition affects dark skin phototypes. The exact pathogenesis of exogenous ochronosis is not completely elucidated. It is thought to be the result of an inhibition of HGD by hydroquinone leading to an oxidation of homogentisic acid and photo-oxidation [3]. Dogliotte and al classified exogenous ochronosis into three clinical stages:

1. Erythema and mild hyperpigmentation
2. Colloid milium lesions and atrophy
3. Papulo-nodular lesions.

The lesions occur particularly over bony prominences such as the temples, forehead, and lower jaws [2]. Dermoscopy shows irregular globular structures that are brown-gray, brown-gray, annular, and arciform, sometimes obliterating the follicular openings. They correspond histologically to ochronotic pigment distributed in the superficial dermis into yellow-brown, sharply defined fragmented fibers [4]. We did not find reports in the literature of exogenous ochronosis occurring in facial lichen planus pigmentosus. Dermoscopy of lichen planus pigmentosus can show dark brown to grey dots and globules of smaller size sparing the follicular openings. They can be distributed in arciform configuration but they are thin not as thick in ochronosis [5]. Various therapeutic agents have been used to treat exogenous ochronosis without complete clearance of the condition. Topical retinoic acid, glycolic acid, and topical corticosteroid were been used with partial improvement. Combined treatment of dermabrasion and CO₂ laser showed improvement of the lesions [3]. The Q-switched alexandrite laser has been reported to be effective to clear ochronotic pigment from the dermis [6].

Another study showed satisfactory results proven histologically with Q switched Nd Yag laser [7].

The particularity of our case is the occurrence of exogenous ochronosis in facial lichen planus pigmentosus and not in melasma lesions. We want also to highlight the usefulness of

dermoscopy in the follow-up of lichen planus pigmentosus since it allowed us to distinguish the two conditions.

Conclusion

We highlight the fact that hydroquinone prescription should be evaluated by practitioners to detect early side effects such as exogenous ochronosis and the contribution of dermoscopy to distinguish lichen planus pigmentosus from exogenous ochronosis.

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