

Vulvar lesion mimicking vitiligo: A case report

Paula Gutierrez^{a,b}, Kushal Gandhi^a, Nnana Amakiri^a, John Garza^{a,b}, Gary Ventolini^{a,*}

^a Texas Tech University Health Sciences Center at the Permian Basin, Odessa, TX, USA

^b The University of Texas Permian Basin, Odessa, TX, USA

ARTICLE INFO

Article history:

Received 8 June 2020

Accepted 12 June 2020

Keywords:

Post-inflammatory hypopigmentation (PIH)

Melanocytes

Skin disorders

ABSTRACT

Background: Post-inflammatory hypopigmentation (PIH) is an acquired partial or total loss of pigment that occurs as a result of cutaneous inflammation. Clinically, post-inflammatory hypopigmentation is recognized by a discoloration of the skin; however, proper diagnosis requires a skin biopsy. Although post-inflammatory hypopigmentation is similar in appearance to vitiligo, histopathological evaluation highlights the key differences in the presence of melanocytes and melanophages.

Case presentation: A 28-year-old woman presented with discoloration in the vulvovaginal area. Physical examination was within normal limits; however, a genital exam revealed a large, intensely white discoloration in an hourglass pattern involving the clitoris, labia majora, and perianal area. Pigmentation was observed at the base of the hair follicles, which is not consistent with vitiligo. The patient consented to a skin biopsy, which was performed without complication. The biopsy showed mild chronic vulvitis and pigment incontinence due to post-inflammatory hypopigmentation. The patient was prescribed a high-potency topical steroid and a significant reduction in lesion area was observed at 3-month follow-up.

Discussion: Proper diagnosis via clinical examination and skin biopsy is essential in the treatment of pigment deficiencies. In cases of post-inflammatory hypopigmentation, the initial cause of inflammation must first be identified in order to provide an effective treatment regimen. When facing uncommon dermatological conditions such as post-inflammatory hypopigmentation, proper histopathological diagnosis, course of treatment, and follow-up are important in order to achieve patient satisfaction.

© 2020 The Authors. Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Melanocytes can react to trauma or cutaneous inflammation with varying levels of melanin production, manifested as hyperpigmentation or hypopigmentation. Post-inflammatory hypopigmentation (PIH) is an acquired partial or total loss of skin pigmentation that occurs after cutaneous inflammation injury. PIH can be caused by skin disorders such as pityriasis lichenoides chronica, atopic dermatitis, and lichen striatus. In addition to various skin disorders, PIH may also be a result of skin damage caused by burns, irritants, and chemical dermatological procedures [1].

There is limited information about the mechanism and pathogenesis of PIH. A major problem is to understand the variation in individual response to inflammation. Clinical presentation of pigment incontinence is recognized by a discoloration of the skin and proper diagnosis requires clinical examination and skin biopsy with histopathological evaluation and confirmation [2].

PIH is often confused with vitiligo, which presents similarly in physical appearance. The main difference is that histological examination shows vitiligo to have an absence of melanocytes, whereas PIH is characterized by melanocytes that exhibit pigment incontinence. In addition, PIH is associated with the presence of melanophages, which are not a feature of vitiligo [1]. It has been observed that PIH is more common among patients with dark skin; there is no gender difference in prevalence [3].

2. Case presentation

A 28-year-old patient consulted a specialist clinic complaining of a discoloration in the vulvovaginal area. The patient had been previously diagnosed with vitiligo by two healthcare providers, but no biopsy had been performed for confirmation. The patient was distressed about her physical appearance, and very concerned that her condition might cause marital issues.

The patient's family history was noncontributory. Her physical exam was otherwise within normal limits. A large, intensely white, and diffuse discoloration in an hourglass pattern, involving the clitoris, labia majora, and perianal area was observed (Fig. 1). At close and magnified observation, in the lateral areas less involved, it was possible to observe

* Corresponding author at: School of Medicine, Texas Tech University Health Sciences Center at the Permian Basin, 800 W. 4th Street, Odessa, TX 79763, USA.

E-mail address: gary.ventolini@ttuhsc.edu (G. Ventolini).



Fig. 1. Post-inflammatory hypopigmentation (PIH) observed at the time of the patient's initial visit.

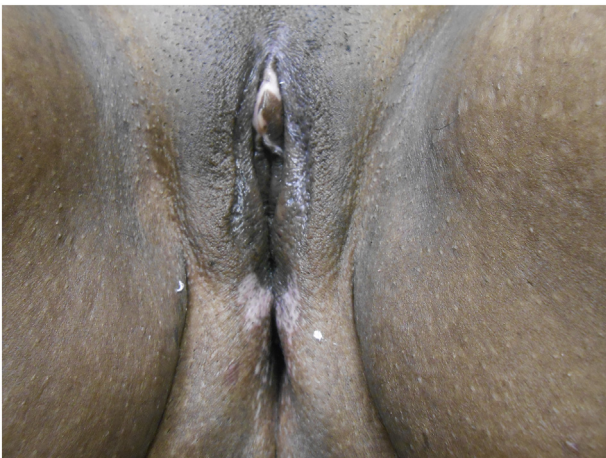


Fig. 2. Reduction of post-inflammatory hypopigmentation (PIH) observed after one round of treatment. Image was obtained 2 months after the initial visit.

that the lesion was composed of white dots that circled the bases of the hair follicles, rather than complete lack of pigment. The patient was reassured and consented to a vulvar biopsy, which was performed in usual fashion without complications.

The pathology report listed a diagnosis of pigment incontinence on a background of mild chronic vulvitis. A Fontana-Masson stain showed preservation of dermal melanocytes. Pigment incontinence was due to post-inflammatory pigment alteration.

With diagnostic confirmation, the patient was prescribed a high-potency topical steroid to be applied to the affected area twice daily for four weeks. At a two-month follow-up, the lesion's appearance had improved (Fig. 2). At a three-month follow-up, the lesion had almost completely resolved, except for a few small areas, including the clitoris.

3. Discussion

Proper diagnosis via biopsy is essential in treating patients with persistent pigment deficiencies. As in the case described, this hypopigmentation can be difficult to cope with. Treatment involves managing the initial inflammatory condition. First-line therapy includes

topical steroids and sun or ultraviolet exposure to aid in re-pigmentation. Topical treatments are typically sufficient for treatment of this post-inflammatory condition and may include application of 1% pimecrolimus, 0.1% 8-methoxypsoralen and/or 0.5–1% coal tar or anthralin. When topical therapy is unsuccessful, chemical peeling and laser therapy may be beneficial. A 308 nm excimer laser may stimulate pigmentation in certain cases of hypopigmentation; however, regular subsequent treatment is necessary to maintain results [1]. When topical ointments have failed to provide desirable effects, some success has been achieved with the use of psoralen plus ultraviolet A photochemotherapy (PUVA) [4].

Proper clinical and pathological diagnosis are required when facing uncommon dermatological conditions. Additionally, adequate treatment and follow-up are necessary for patient satisfaction and reassurance.

Contributors

Paula Gutierrez contributed to the literature search and to writing and editing the manuscript.

Kushal Gandhi contributed to the literature search and to writing and editing the manuscript.

Nnana Amakiri contributed to the literature search and to writing and editing the manuscript.

John Garza contributed to the literature search and to writing and editing the manuscript.

Gary Ventolini consulted, obtained consent, and treated the patient.

Conflict of interest

The authors declare that they have no conflict of interest regarding the publishing of this document.

Funding

No funding from an external source supported the publication of this case report.

Patient Consent

Obtained.

Provenance and Peer Review

This case report was peer reviewed.

Acknowledgements

We are grateful to the Texas Tech University Health Sciences Center, Permian Basin, TX, and Ailena Mulkey, Evangelina Santiago, and Jammie Holland from Clinical Research Institute (CRI), TX for their excellent support and help on this study.

References

- [1] V. Vachiramon, Thadanipon K. Postinflammatory hypopigmentation, *Clin. Exp. Dermatol.* 36 (7) (2011) 708–714.
- [2] L. Nieuweboer-Krobotova, Hyperpigmentation: types, diagnostics and targeted treatment options, *J. Eur. Acad. Dermatol. Venereol.* 27 (Suppl. 1) (2013) 2–4.
- [3] R.M. Halder, P.E. Grimes, C.I. McLaurin, M.A. Kress, J.A. Kenney Jr., Incidence of common dermatoses in a predominantly black dermatologic practice, *Cutis.* 32 (4) (1983) 388(90).
- [4] I. Lopez, A. Ahmed, A.G. Pandya, Topical PUVA for post-inflammatory hypopigmentation, *J. Eur. Acad. Dermatol. Venereol.* 25 (6) (2011) 742–743.