Case Reports in Dermatology

Case Rep Dermatol 2021;13:58–61

DOI: 10.1159/000511695
Published online: February 1, 2021

Single Case

Eosinophilic Vulvar Leukoderma

Kaya Nakada[a] Shiro Niiyama[a] Toshiaki Oharaseki[b] Hidetsugu Fukuda[a]

[a]Department of Dermatology, Toho University Ohashi Medical Center, Tokyo, Japan;
[b]Department of Pathology, Toho University Ohashi Medical Center, Tokyo, Japan

Keywords
Atopic dermatitis · Eosinophilic esophagitis · Lichen sclerosis

Abstract
We report the case of a 54-year-old woman with asthma and atopic dermatitis who presented a white spot on the genitalia. Histologic examination showed numerous eosinophils in the epithelium and the dermis. Eosinophilic esophagitis is defined as an esophageal disease characterized clinically by symptoms related to esophageal dysfunction and histologically by an eosinophil-predominant inflammation. We discuss the possible relationship between the two diseases.

Introduction

We present an atopic dermatitis (AD) patient with eosinophilic vulvar leukoderma and discuss this case in relation to eosinophilic esophagitis (EoE).

Case Report

A 54-year-old woman presented with a 1-year history of white spot on the genitalia. She had a history of asthma and AD and was taking budesonide/formoterol fumarate hydrate,

Shiro Niiyama
Department of Dermatology
Toho University Ohashi Medical Center
2-22-36 Ohashi, Meguro-ku, Tokyo 153-8515 (Japan)
sniiyama@aol.com

montelukast sodium, and topical corticoids. Physical examination revealed leukoderma at the vestibule and extending into the introitus. No eczematous change was noted (Fig. 1a). No leukoderma was noted in the oral mucosa. The white blood cell count was 6.4 × 10^9/L with 4.5% eosinophils, increased level of immunoglobulin E (5,271.0 IU/mL), and other routine biochemical tests were normal. Histologic examination showed dilation of intercellular spaces and eosinophils in the epithelium, and numerous eosinophils (eosinophil count >15 in a high-power field) and lymphohistiocytes in the dermis (Fig. 1b, c). The Fontana-Masson stain and periodic acid-Schiff stain were negative. Endoscopic examination performed to check for concurrent EoE revealed findings suggestive of esophagitis, although histopathology revealed no eosinophilic infiltration of the organ. The patient has been under topical dexamethasone treatment for 3 months, and the leukoderma remains unchanged (Fig. 2a). In a follow-up biopsy, the features of spongiosis and eosinophilic infiltration were absent (Fig. 2b, c).

Discussion

The differential diagnosis for vulvar leukoderma in this case includes lichen sclerosus, lichen planus, leukoplakia, and vitiligo. Histologic findings of lichen sclerosus show epidermal atrophy, hydropic basal degeneration, and upper papillary dermal pallor with a band of lymphocytic infiltrates. Neither epidermal atrophy nor hydropic basal degeneration was seen in the present case, and eosinophilic infiltration is not encountered in lichen sclerosus. Lichen planus produces a triad of histologic features that include hyperkeratosis, basal layer destruction, and a lichenoid infiltrate at the dermal-epidermal junction. None of these histologic features was noted in this case. The biopsy in our case lacked histologic evidence of nuclear crowding, cellular atypia, and increased mitotic index seen in vulvar leukoplakia. In the present case, the Fontana-Masson staining yielded a negative result; hence, the lesion did not contain melanin, similar to the case with vitiligo; however, eosinophilic infiltration is not encountered in vitiligo. Leukoderma may be seen in some instances in the form of postinflammatory depigmentation in AD; however, this is usually not seen in the vulvovaginal region [1]. Eosinophilic leukoderma is a characteristic feature in the present case.

EoE is a new disease defined as “an esophageal disease characterized clinically by symptoms related to esophageal dysfunction and histologically by an eosinophil-predominant inflammation” [2]. It is caused by a T-helper type 2 cell response to food antigens in contact with the esophageal mucosa in patients with atopic conditions [2]. Prevalence of AD in EoE is 46.1% [3]. One of the characteristic endoscopic findings is white spots and 15 or more eosinophils in at least a high-power field within these biopsies [4]. The most effective treatment of EoE is systemic or topical corticosteroids [2]. There has been no published paper in the field of dermatology dealing with eosinophilic leukoderma, while EoE presents with findings remarkably similar to those noted in this case. Although the present case had no concurrent EoE at this point, further accumulation of cases is needed to clarify the association between these two disorders.

Statement of Ethics

The patient has given her informed consent for the publication of this case. Research was conducted ethically in accordance with the World Medical Association Declaration of Helsinki.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Funding Sources

No funding was received.

Author Contributions

K.N. was involved in direct management of the patient. S.N. drafted the first manuscript and reviewed the literature. T.O. and H.F. supervised the manuscript drafting. All authors read and approved the final manuscript.

References

1 Kuriyama S, Kasuya A, Fujiyama T, Tatsuno K, Sakabe J, Yamaguchi H, et al. Leukoderma in patients with atopic dermatitis. J Dermatol. 2015 Feb;42(2):215–8.
2 Straumann A, Katzka DA. Diagnosis and treatment of eosinophilic esophagitis. Gastroenterology. 2018 Jan;154(2):346–59.
3 Mohammad AA, Wu SZ, Ibrahim O, Bena J, Rizk M, Piliang M, et al. Prevalence of atopic comorbidities in eosinophilic esophagitis: A case-control study of 449 patients. J Am Acad Dermatol. 2017 Mar;76(3):559–60.
4 Liacouras CA, Furuta GT, Hirano I, Atkins D, Attwood SE, Bonis PA, et al. Eosinophilic esophagitis: updated consensus recommendations for children and adults. J Allergy Clin Immunol. 2011 Jul;128(1):3–20.e6.

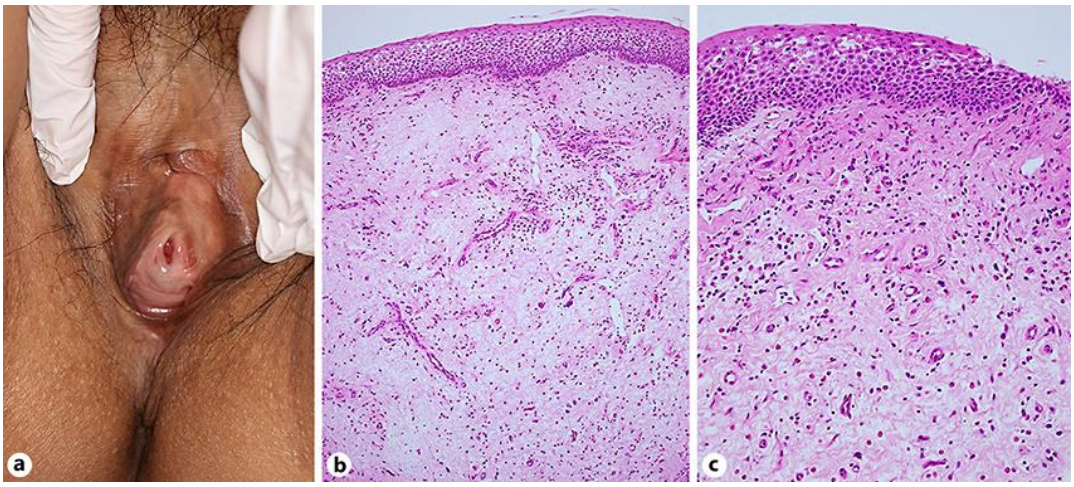

Fig. 1. **a** Leukoderma at the vestibule and extending into the introitus. Dilation of intercellular spaces and eosinophils in the epithelium, and numerous eosinophils in the dermis. H&E stain: ×100 (**b**), ×200 (**c**).

Case Rep Dermatol 2021;13:58–61
DOI: 10.1159/000511695

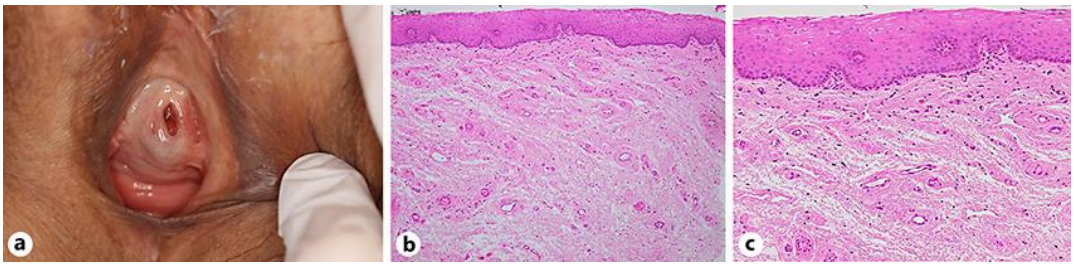

Fig. 2. **a** Leukoderma remains unchanged after topical dexamethasone therapy. The features of spongiosis and eosinophilic infiltration had disappeared. H&E stain: ×100 (**b**), ×200 (**c**).