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**Single Case**

# Primary Cutaneous Marginal Zone Lymphoma following Repeated Inflammation Caused by Hair Dyeing

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**Abstract**
Primary cutaneous marginal zone lymphoma (PCMZL) is a rare form of B-cell lymphoma that primarily affects the skin. Chronic antigen stimulation has been implicated in its development, with cases associated with various triggers. We present a case of PCMZL following chronic inflammation caused by long-term hair dyeing. A 75-year-old woman with a history of repeated inflammation and itching after hair dyeing for 30 years presented with persistent red-to-violaceous patches and plaques on her scalp. Despite receiving topical corticosteroid treatment for 10 years, the lesions remained. Pathological examinations confirmed the diagnosis of PCMZL. The patient achieved complete remission after radiotherapy. This case underscores the potential link between chronic inflammation and the development of PCMZL.



## Introduction

Primary cutaneous marginal zone lymphoma (PCMZL), a subtype of primary cutaneous B-cell lymphoma, is a rare, indolent B-cell lymphoma without evidence of extracutaneous disease. Skin manifestations include violaceous papules, plaques, and nodules that occur in multiple locations, often with no symptoms. Chronic antigen stimulation is suggested to play a crucial role in the development of PCMZL, with reports of cases caused by insect bites, tattoos, or vaccination [1, 2]. Herein, we report a case of PCMZL following a history of chronic inflammation caused by long-term hair dyeing.

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## Case Report

A 75-year-old woman visited our clinic with red-to-violaceous patches and plaques on her scalp that had appeared 10 years prior (Fig. 1a, b). She was taking medication for hypertension, and no other underlying diseases were stated. She described repeated inflammation with erythema and itching after hair dyeing for 30 years. For 10 years, she underwent intermittent oral and topical corticosteroid treatment using antihistamines, without improvement. Dermoscopic examination revealed a red-to-purple background with serpentine arborizing vessels (Fig. 1c). Pathological examination via punch biopsy revealed dense atypical lymphoid cell infiltration in the dermis under H&E staining (Fig. 2a) and positive CD20 cells (Fig. 2b) and focal positive Bcl-2 and CD3 cells (Fig. 2c, d) by immunohistochemical staining. CD10-positive cells were not observed (Fig. 2e). F-18 FDG PET examination was performed to rule out the involvement of other organs. Mild hypermetabolic skin lesions on the scalp were disclosed, but no involvement of other organs was observed. The patient was diagnosed with PCMZL, and complete remission was achieved after 18 cycles of radiotherapy using 2 Gy.

## Discussion

PCMZL is an indolent B-cell lymphoma limited to the skin. Histological examination may reveal infiltration by small lymphocytes, marginal zone B-cells, lymphoplasmacytoid cells, and plasma cells. CD20, CD79a, and Bcl-2 appear to be positive, while CD10 and Bcl-6 are usually negative, owing to their B-cell lineage. Solitary lesions can be effectively treated using radiotherapy or surgical excision, while multifocal lesions can be treated with rituximab or chlorambucil therapy. If no extracutaneous infestation is present, the 5-year survival rate is as high as 99%, representing an indolent disease course [3].

The precise pathogenesis of PCMZL is not yet well known, but chronic antigen stimulation is suggested to play a key role in its development [4]. In case reports, some preceding factors such as vaccination, radiation therapy, tattoos, or tick bites have been suggested as potential antigenic stimulations [1, 2]. In this case, the patient had a history of repeated inflammation on her scalp due to hair dyeing, which could have been the chronic antigen stimulation that caused PCMZL. P-Phenylenediamine, an organic compound widely used in hair dyes, is a potent contact sensitizer. Relatively high and stable prevalence rates of contact allergies to P-phenylenediamine have been observed in European patients with dermatitis [5]. In conclusion, we report a case of PCMZL diagnosed after a 30-year history of chronic inflammation caused by hair dyeing, which was successfully treated using radiotherapy. Therefore, chronic antigen stimulation may play a crucial role in the pathogenesis of PCMZL.

Our case report has certain limitations that should be acknowledged. First, we did not include immunohistochemical markers such as CD5 or IRTA-1 in our analysis. Second, the assessment of chronic inflammation was solely based on the patient's description as there were no outpatient visits recorded over the 30-year period. Lastly, despite our thorough investigation, we were unable to provide a definitive explanation for the exact pathogenesis of disease occurrence. These limitations highlight the importance of conducting further research in the future to address these gaps in knowledge. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000533516).

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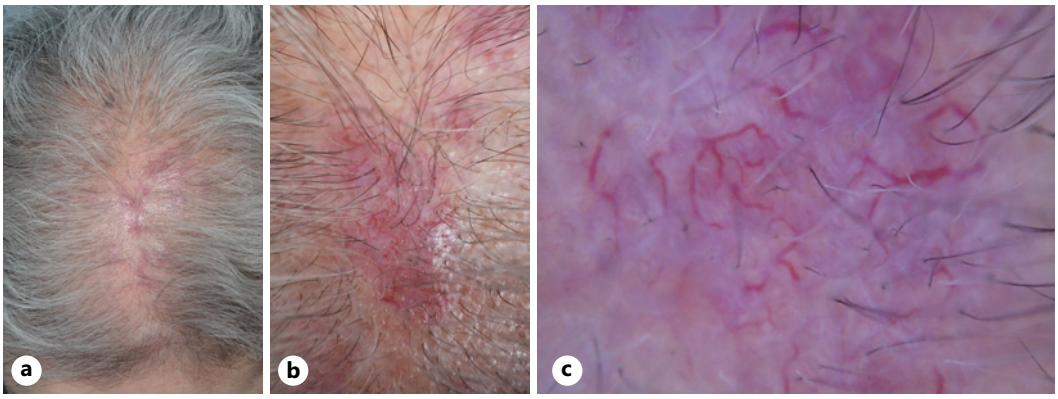


**Fig. 1. a**, **b** Multiple red-to-violaceous patches and plaques on the scalp. The patient claimed itchiness was the only symptom. **c** Dermoscopic findings showed serpentine arborizing vessels in a red to purple background.

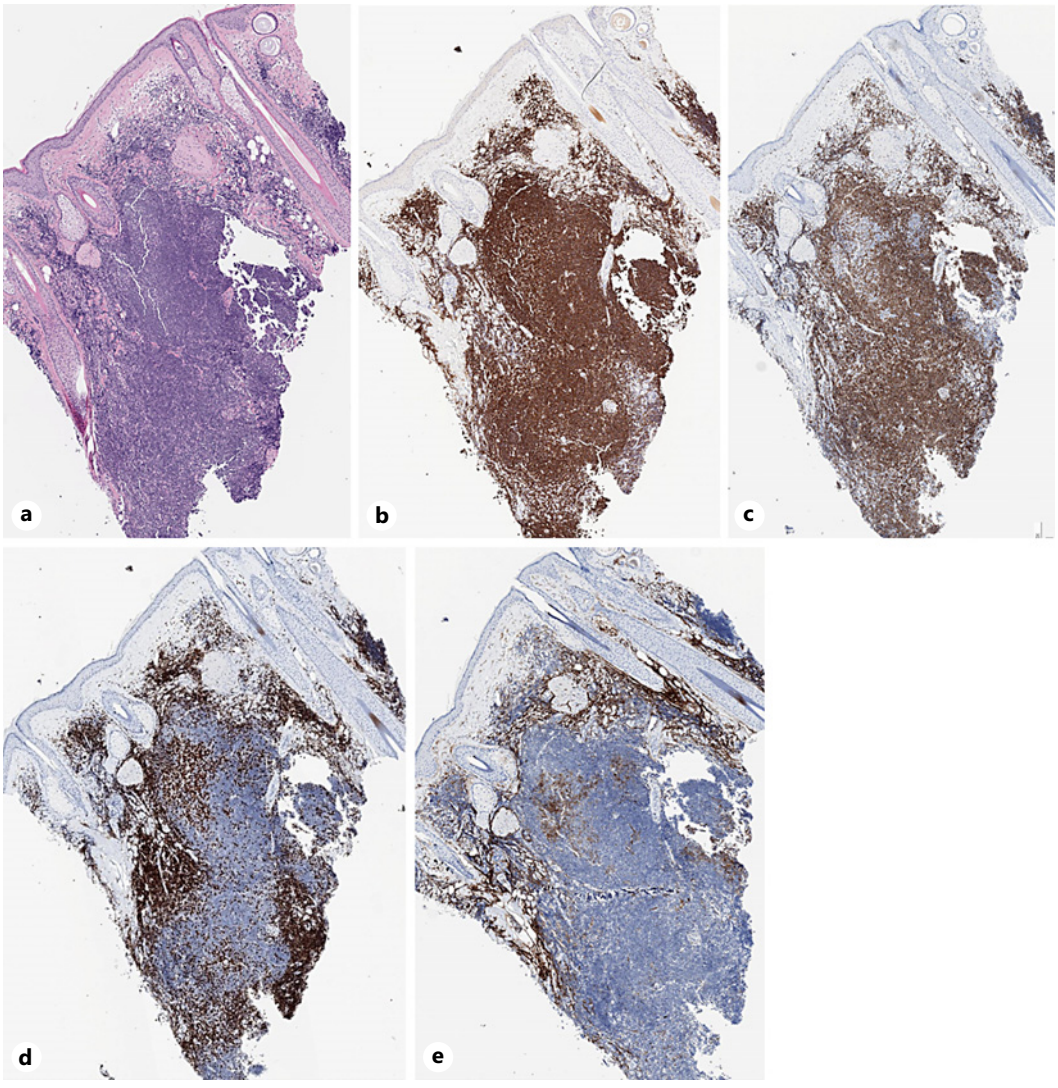


**Fig. 2. a** H&E staining of punch biopsy specimen showed dense atypical lymphoid cell infiltration in the dermis. **b**–**e** Immunohistochemical staining revealed CD 20-positive (**b**) and focal Bcl-2-, CD3-positive cells (**c**, **d**) and no CD 10-positive cells (**e**). **a**–**e**: ×40.

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Author Ui Hyeon Jo was not available to confirm co-authorship, but the corresponding author Dae Hun Suh affirms that author Ui Hyeon Jo contributed to the paper, had the opportunity to review the final version to be published, and guarantees author Ui Hyeon Jo co-authorship status and the accuracy of the author contribution and conflict of interest statements.

## Statement of Ethics

Ethical approval is not required for this study in accordance with local or national guidelines. Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images.

## Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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## Author Contributions

The data were collected and the initial manuscript was written by Jun Hyo Lee, Ui Hyeon Jo, and Tae Min Kim. Dae Hun Suh assessed and edited the manuscript, offering valuable feedback and contributing to the final version. Tae Min Kim and Dae Hun Suh provided medical treatment to the patient. The ultimate version of the manuscript was authored by Jun Hyo Lee.

## Data Availability Statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

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