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Lichen Planus Induced by Stevens Johnson Syndrome

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

The present study reports the appearance of lichen lesions with genital and nail involvement 2 months after the onset of a Stevens-Johnson syndrome (SJS) associated with lamotrigine use. A chronic inflammatory disorder called lichen planus (LP) most frequently affects middle-aged adults. LP can affect the skin or mucous membranes, such as the conjunctiva, vulvovaginal, esophageal, and laryngeal mucosa. A 23-year-old patient was hospitalized in dermatology for SJS that occurred 9 days after taking lamotrigine for treatment of severe depression. The occurrence of cutaneous lichen lesions on the site of a healed dermatosis has been widely reported under the name of Wolf's isotopic response. This isotopic response refers to the appearance of a new dermatosis on the site of an old healed dermatosis.

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1. INTRODUCTION

Lichen planus (LP) is a chronic inflammatory disorder that most often affects middle-aged adults. LP can involve the skin or mucous membranes including the oral, vulvovaginal, esophageal, laryngeal, and conjunctival mucosa [1]. Its etiology is unknown. Although several drugs have been implicated in inducing lichen planus [2], a case of lichen cause by Steven Johnson syndrome (SJS) has never been reported in the literature.

We report a case of lichen induced by Steven Johnson syndrome due to lamotrigine.

2. CASE REPORT

A 23-year-old patient was hospitalized in dermatology for Steven Johnson syndrome (SJS) that occurred 9 days after taking lamotrigine for treatment of severe depression. On clinical examination, there were pseudo-bullae on the entire skin, the Nikolsky sign was negative, with involvement of the oral and genital mucosa of the small lips and without phanerian involvement (Figs. 1,2). A cutaneous biopsy was in favor of a toxidermia type Steven Johnson syndrome. Complementary examinations found an inflammatory syndrome; renal and liver function were normal. The evolution was marked by the healing of cutaneous and mucosal lesions from the fifteenth day after stopping the medication and local care. 2 months after the patient reported significant genital itching. Clinical examination showed lichenification of the labia majora. The rest of the somatic examination found onychodystrophy and trachyonychia in 10 nails of the hands, the hyperchromic scars of the entire skin were uninvolved (Figs. 3,4). Indication for the performance of an ungual biopsy was made but due to the patient's refusal, the histology of a genital cutaneous biopsy showed orthokeratotic hyperkeratosis, hypergranulosis, irregular hyperacanthosis with a dermal lymphocytic infiltrate, leading to a conclusion of lichen planus (Fig. 5). The patient was treated with betamethasone dipropionate and disodic phosphate with an intramuscular injection of 0.5 mL/month for 9 months. The evolution was good, marked by the disappearance of lichenification and genital itching as well as improvement of nail involvement.

3. DISCUSSION

Our observation reports the appearance of lichen lesions with genital and nail involvement 2 months after the onset of a Stevens-Johnson syndrome (SJS) associated with lamotrigine use.

Stevens-Johnson syndrome or toxic epidermal necrolysis is a rare and severe medication-induced skin reaction characterized by the massive death of keratinocytes resulting from the activation of the immune system and induced by different signals combining the production of cytolytic proteins by CD8 cytotoxic T cells and the activation of apoptotic or necrotic pathways in keratinocytes by different soluble mediators [3]. Regarding lichen planus, its exact cause has not been determined but the pathogenesis by keratinocyte apoptosis induced by TNF alpha released by memory T CD8 lymphocytes is well described. These activated memory phenotype T CD8+ lymphocytes are preferentially found next to damaged basal keratinocytes. This could potentially explain the involvement of Stevens-Johnson syndrome in the development of lichen planus in our patient [4].

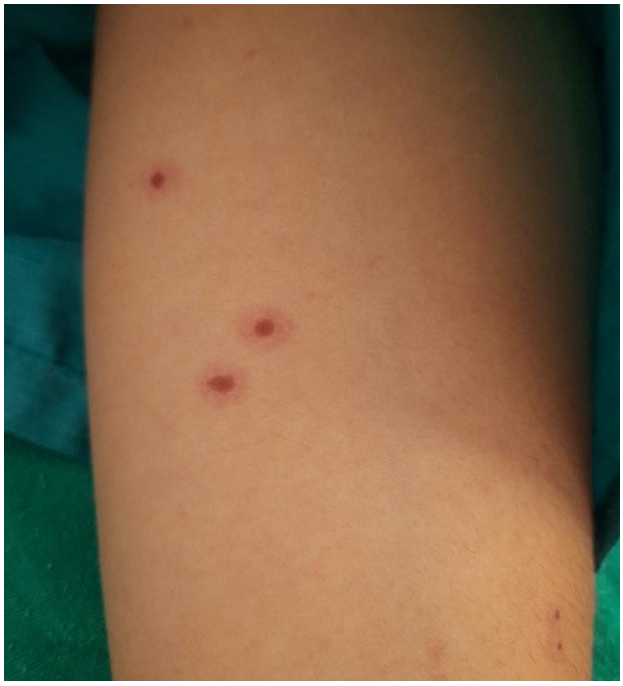
Also in the pathophysiology, in a recent study, Ziemer and al found that the epidermal keratinocytes of all SJS/TEN cases showed programmed cell death protein 1 (PD-L1) expression on the cell surface, in contrast to lichen planus, where epidermal expression was absent and significant expression was instead seen in the lymphocytic inflammatory infiltrate [5].

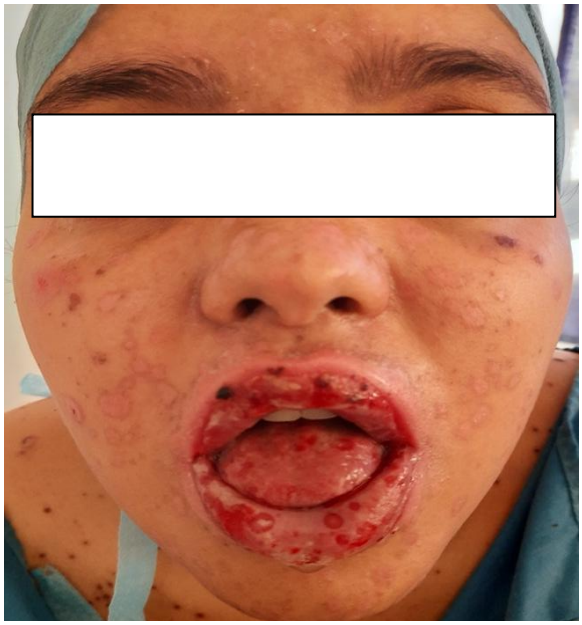
The originality of this presentation lies in the fact that the nail lichen lesions developed after the onset of SJS and spared the old scars, which to our knowledge has never been described in the literature.

Unlike our case, Saka et al. described the appearance of cutaneous lichen lesions on the scars of post-bubble ulcerations of SJS [6]. The occurrence of cutaneous lichen lesions on the site of a healed dermatosis has been widely reported under the name of Wolf's isotopic response. This isotopic response refers to the appearance of a new dermatosis on the site of an old healed dermatosis. The first dermatosis is in the majority of cases, a herpes viral infection, mostly shingles, rarely chickenpox, herpes, minor polymorphic erythema, or vitiligo [7,8]. The dermatoses that develop secondarily are mostly

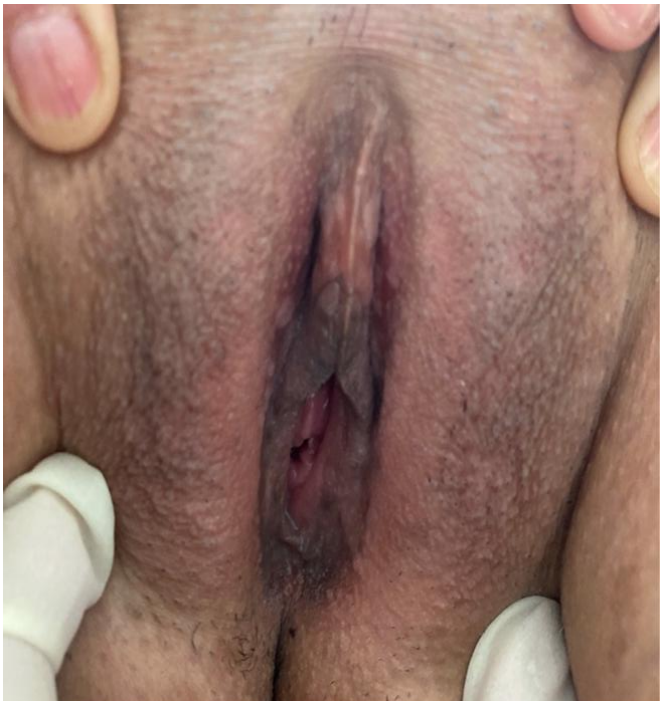
tumoral and granulomatous, with others being infectious (secondary syphilis, molluscum contagiosum), inflammatory and dysimmune (rosacea, sarcoidosis, psoriasis, morphea, lichen planus, sclerosing lichen, lupus, pemphigoid [9].

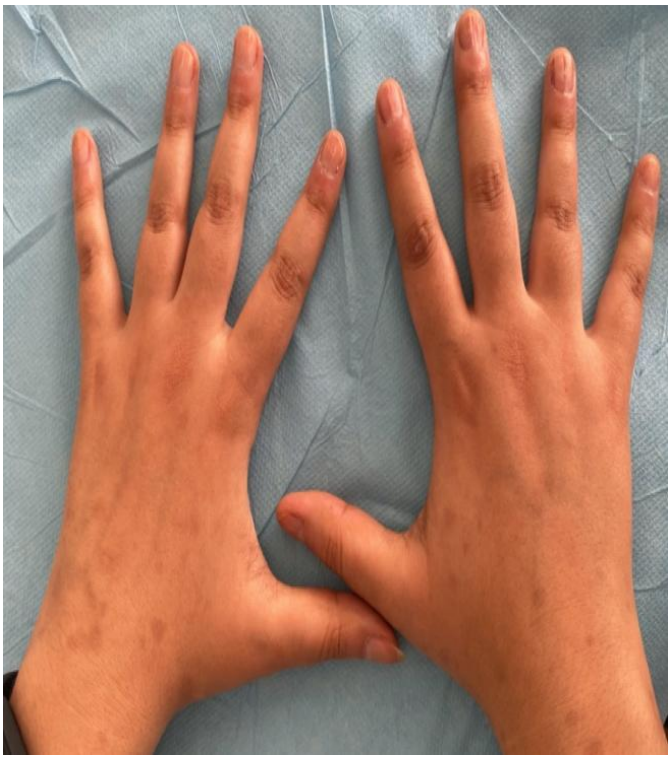
Another case was described by F. Worsnop and colleagues, in which a 32-year-old woman developed toxic epidermal necrolysis secondary to sulfasalazine, treated with infliximab. Infliximab treatment subsequently triggered erosive lichen planus (LP) involving the mouth and vulva [10], but our patient received no treatment for her SJS.





Figs. 1 and 2. Mucocutaneous lesions of Steven Johnson syndrome
(1: Three pseudo blackouts on the forearm. 2: Involvement of the oral and conjunctival mucosa.)





Figs. 3 and 4. Lesions of the genital mucosa and nail of lichen (3: Lichenification of the labia majora; 4: Onychodystrophy and trachyonychia of the 10 fingernails)

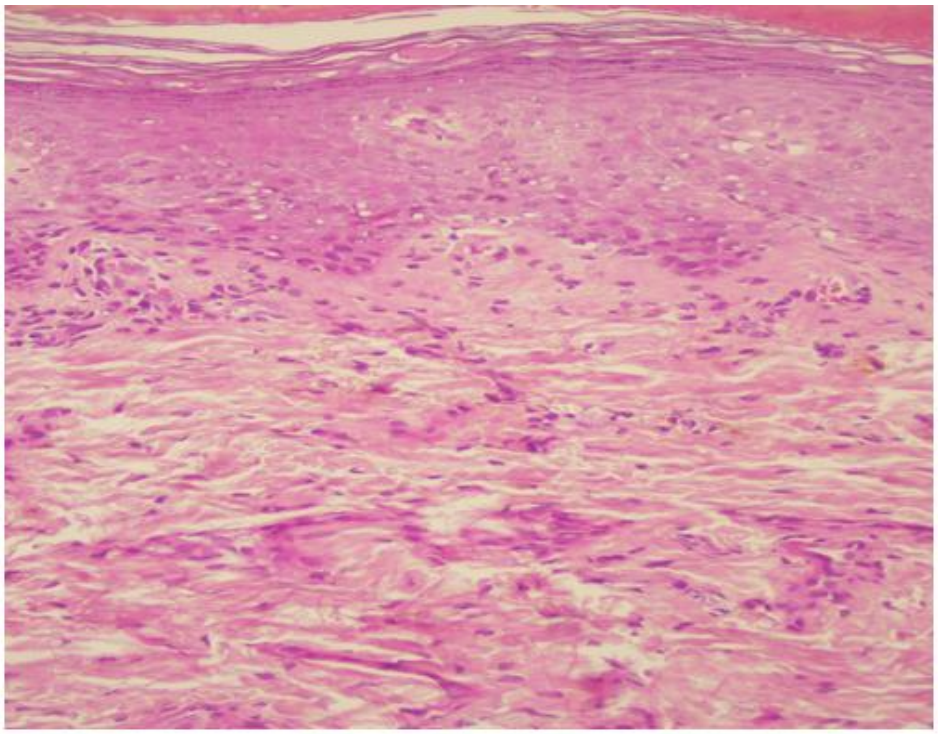


Fig. 5. Histology of a genital cutaneous biopsy showing orthokeratotic hyperkeratosis, hypergranulosis, irregular hyperacanthosis with a dermal lymphocytic infiltrate, leading to a conclusion of lichen planus

4. CONCLUSION

We report the first case of lichen planus induced by Stevens Johnson syndrome. The occurrence of these two skin disorders can be explained by their pathophysiological mechanisms involving memory TCD8 lymphocytes.

CONSENT STATEMENT

The patient has given informed consent prior to inclusion in the case report.

ETHICAL APPROVAL

The authors have obtained all necessary ethical approval from suitable Institutional or State or National or International Committee

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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