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Single Case

A Case of Lip Psoriasis in a 14-Year-Old Boy Successfully Treated with Adalimumab

Francesca Caroppo[a] Laura Gnesotto[a] Maria Ludovica Deotto[a]
Roberto Salmaso[b] Anna Belloni Fortina[a]

[a]Department of Medicine DIMED & Department of Women's and Children's Health SDB, Pediatric Dermatology Regional Center, University of Padova, Padova, Italy; [b]Department of Medicine DIMED, Surgical Pathology and Cytopathology Unit, University of Padova, Padova, Italy

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Abstract
Psoriasis is a common chronic skin disease mainly located in areas of friction. Psoriasis of the lips as an exclusive presentation is rare and often misdiagnosed. Different anti-psoriatic therapies have been proposed, but the literature is limited to case studies with partial results. Biologic therapies have revolutionized the management of many dermatologic conditions, including psoriasis, and they are approved for pediatric use. We report the case of a 14-year-old boy with a 2-year history of white-yellowish scaling lesions on his lips, without intraoral involvement. Lip biopsy showed a psoriasiform pattern. Treatment with adalimumab 40 mg every other week was started, and after 6 months of therapy, we obtained a complete remission of the patient's lip psoriasis.

Introduction

Psoriasis is a chronic inflammatory disorder with a multifactorial etiology, which comprises genetic, immunological, and environmental factors. Several clinical phenotypes of psoriasis are recognized, and typical skin lesions appear as erythematous-desquamative plaques, mainly distributed symmetrically on the scalp, elbows, knees, and lumbosacral area [1]. Psoriasis involvement of the lips is a very uncommon presentation, with only few cases

Correspondence to:
Francesca Caroppo, francesca.caroppo@outlook.it

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reported in the literature [2]. Owing to the infrequency of reports as well as the complexity of labial psoriasis, this manifestation is associated with several differential diagnoses and could be difficult to treat. The introduction of biological therapy has revolutionized the management of many chronic diseases, including psoriasis [3]. We report a rare case of isolated lip psoriasis in a 14-year-old boy successfully treated with adalimumab, after the failure of conventional therapies. 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/10.1159/000532103).

Case Report

A 14-year-old boy presented to our Pediatric Dermatology Unit for the appearance of white-yellowish scaling lesions on his lips, which occurred 2 years before and worsened in the last 6 months. The patient referred burning sensations and pain, especially during eating and when exposed to cold, and he also complained about intense esthetic discomfort. Physical examination revealed the presence of white-yellowish thick scales, easily detachable from both the upper and lower lips, revealing an erythematous background and fissuring (shown in Fig. 1). There was neither involvement of the inner mucosa nor the surrounding skin. The patient reported the absence of familial or personal history of psoriasis, atopic dermatitis, or any other dermatological conditions. Moreover, he denied any history of contact with topical antibiotics or cosmetics. Routine laboratory examinations were normal, including total immunoglobulin E and anti-transglutaminase antibodies, as well as the patch test. Emollient medications, topical steroids, and antifungal and antiviral creams had previously been prescribed without benefit. Because of the lesions' chronic nature and the failure of different therapies, an incisional biopsy was performed from the lower lip. Histological examination revealed epidermal acanthosis with elongated rete ridges, and a mild lymphocytic infiltrate in the superficial dermis (shown in Fig. 2). Based on histological findings and clinical presentation, a diagnosis of isolated lip psoriasis was postulated. Eighteen narrowband ultraviolet B phototherapy sessions were prescribed to our patient, without improvement. Considering the failure of conventional therapies, adalimumab 40 mg every other week was introduced. After 2 months of therapy, the patient presented significant improvement in his clinical condition, showing good resolution of scaling lesions (shown in Fig. 3a). After 6 months, the situation further ameliorated with complete remission of psoriasis and great satisfaction of the patient. Only mild lip xerosis remained, which was treated with a topical emollient (shown in Fig. 3b).

Discussion

Psoriasis is a common inflammatory, immune-mediated, chronic skin disorder, which may seldom present on buccal mucosa, tongue, gingiva, palate, and very rarely on lips and perioral area [4]. Possible clinical presentations in these sites comprise white or grayish-yellow scales, ulcerative, vesicular, pustular, and indurated entities as well as diffuse mucosal erythema [5]. Previous studies showed that lip psoriasis could develop independently of, concurrently with, or prior to the onset of typical cutaneous manifestations [6]. Since 2000, only 15 cases of lip psoriasis have been reported in the literature until now and 2 of these involved pediatric patients (aged ≤18 years) [2]. Migliari et al. [7] described the only case with intraoral involvement, regarding a 13-year-old girl treated with vitamin A derivatives and topical steroids without effect. The second pediatric report, by Sehgal et al. [8], concerned a female of age

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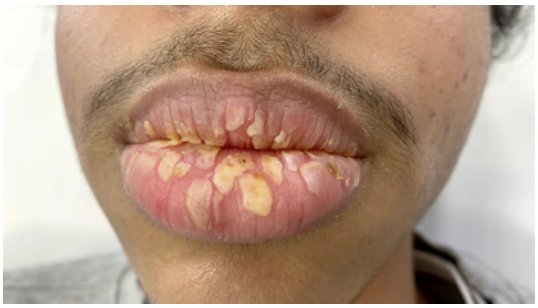

Fig. 1. At baseline visit, the patient presented white-yellowish thick scales, easily detachable from both the upper and lower lips.

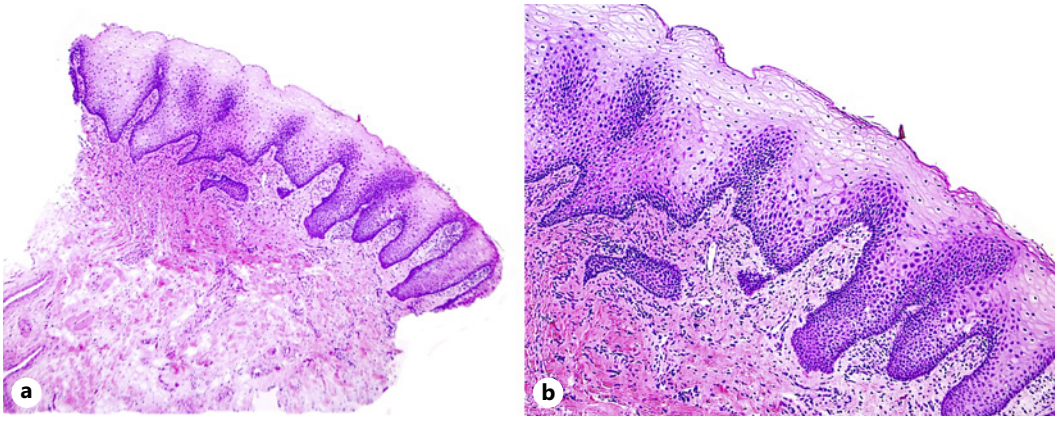

Fig. 2. Histological sections of lip biopsy (stained with hematoxylin and eosin [H&E]) under ×4 (**a**) and ×20 (**b**) magnifications. Histological examination showed epidermal acanthosis with elongated rete ridges and a mild lymphocytic infiltrate in the superficial dermis.

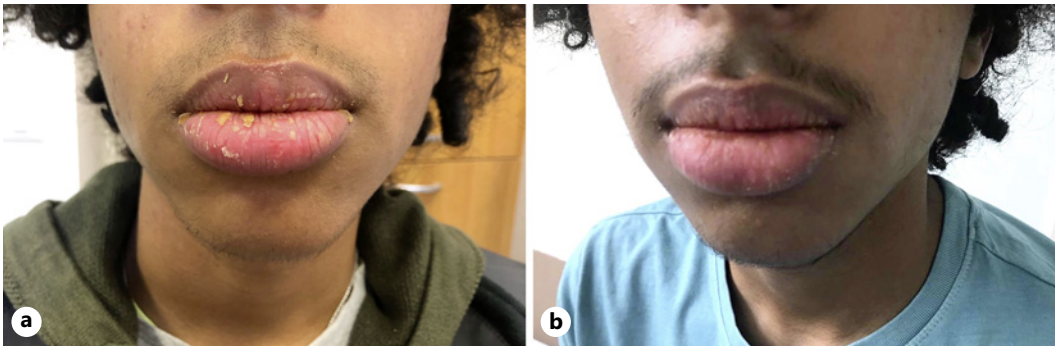

Fig. 3. a After 2 months of therapy. **b** After 6 months of therapy.

16 years who presented with layers of thick, grayish-white scales on her lips. The patient applied topical tacrolimus 0.1% ointment, calcipotriol hydrate, and dipropionate betamethasone with complete remission. Diagnosis of lip psoriasis first requires an accurate clinical examination, searching for other psoriatic lesions on skin, mucosae, and nails. Dermoscopy could be a valid tool since it is a noninvasive technique: psoriatic lesions show a distinctive pattern made up of diffuse white scales with symmetrically and regularly spaced dot vessels, on an erythematous background [9]. It was proposed that mild trauma, chronic irritation, or protruding teeth can lead to psoriatic lesions on the lips, especially in a genetically predisposed individual [6]. Exclusion of other causes is important, particularly if cutaneous

lesions are absent. Lip psoriasis may be clinically confused with cheilitis, chronic eczema, actinic dermatitis, chronic candidiasis, and leucoplakia [10]. There are no standardized treatment protocols or guidelines for affected individuals. Several treatments have been proposed in the literature: topical strong to very strong topical steroids, calcipotriol, tacrolimus, vitamin A derivatives, salicylic acid, and methotrexate with positive results in most cases [2]. Currently, biological drugs appear to be the only systemic medication, besides phototherapy, suitable for pediatric use since they are considered effective and safe. Five biological agents have been approved until nowadays for pediatric psoriasis (adalimumab, etanercept, ustekinumab, secukinumab, ixekizumab) [3, 11]. Adalimumab is a fully human anti-tumor necrosis factor-alpha monoclonal antibody, authorized in Europe as first-line systemic therapy for the treatment of psoriasis in children (aged ≥4 years) who did not respond to topical therapies and phototherapy [11]. Therefore, we decided to treat our patient with adalimumab 40 mg every other week. This therapy proved to be effective already after the first 2 months. Six months later, we achieved a complete remission of his lip lesions. No side effects occurred, and blood tests were normal. To the best of our knowledge, adalimumab has never been reported as a treatment for lip psoriasis in a child. In countries where biological drugs are not approved for the treatment of psoriasis in pediatric age, besides topical corticosteroids, topical calcineurin inhibitors may be also used with high profile of efficacy and safety, especially for the treatment of sensitive areas (such as face and genital area, which are often involved in children with psoriasis). Further studies are necessary in order to assess the long-term efficacy of biological drugs for lip psoriasis and also to evaluate the use of the more recent anti-interleukin agents to treat this specific disorder.

Statement of Ethics

Parents of the patient referred in this manuscript gave their written informed consent to the publication of their son's photographs and medical information. Ethical approval is not required for this study in accordance with national guidelines.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

F.C., L.G., M.L.D., R.S., and A.B.F. have made substantial contributions to conception and design of the study, acquisition of data, analysis, and interpretation of data. F.C., L.G., M.L.D., R.S., and A.B.F. have been involved in drafting the manuscript and revising it critically for important intellectual content. F.C., L.G., M.L.D., R.S., and A.B.F. read and approved the final manuscript.

Data Availability Statement

Data sharing is not applicable to this article as no new data were created or analyzed in this study. Further inquiries can be directed to the corresponding author.

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