Ustekinumab-induced amelioration of both palmoplantar psoriasis and psoriatic glossitis

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INTRODUCTION

Psoriasis is a chronic inflammatory skin disease that may affect any body area, including sensitive sites such as face, nails, scalp, hands, palms, feet, and genitals. Although less frequent, severe forms of either geographic or fissured tongue accounting for psoriasis manifestations can also occur. The involvement of these sites may lead to a marked impairment of quality of life, interfering with work productivity, leisure, and social activities.[1] According to current guidelines, the therapeutic approach for psoriasis of these particular sites, although localized and classified as mild or mild-to-moderate, might require systemic therapies, including biologics.[2]

Abbreviations used:

DLQI: Dermatology Life Quality Index
PASI: Psoriasis Area Severity Index
QoL: quality of life

CASE REPORT

A 16-year-old girl, affected by psoriasis since the age of 4 with a family history for psoriasis, came to our ambulatory care because of the occurrence of psoriatic lesions localized at palmo-plantar areas, trunk, perioral region, and tongue (overall Psoriasis Area Severity Index [PASI] score, 5). In particular, her tongue was swollen, fissured, painful (PAIN-Visual Analog Scale score, 7), and associated with moderate burning sensation and discomfort (Fig 1, *A* and *B*). Because she was a high school student and tennis player, palmo-plantar localization together with tongue involvement had an extremely negative impact on both her social life and sports performances (Dermatology Life Quality Index, 19). The patient was previously treated with topical medications, phototherapy (narrow-band ultraviolet B), and, in 2010, with cyclosporine, which was withdrawn after 4 months because of the occurrence of adverse events (namely, headache). However, none of these therapies, including cyclosporine, obtained satisfactory clinical response both on the palmoplantar lesions and the fissured tongue. Considering the involvement of sensitive areas (ie, tongue, palms, and soles), the high impact on quality of life (QoL), the relative contraindication (teratogenicity and menstrual dysfunctions) of other conventional systemic therapies (ie, methotrexate and acitretin), and parents' reluctancy to initiate a concomitant contraceptive treatment, we decided to start ustekinumab therapy at a 45-mg dose administered according to labeled indications at week 0, week 4, and every 12 weeks thereafter. After 4 weeks, PASI score decreased (PASI score, 3.2), with partial improvement of tongue involvement and related symptoms. After 16 weeks of treatment, the patient achieved complete clearance of all mucocutaneous manifestations, including palmo-plantar lesions, and remission of psoriatic glossitis (PASI score, 0) (Fig 2, *A* and *B*) that was associated with a likewise important QoL improvement (Dermatology Life Quality Index, 0; PAIN-Visual Analog Scale, 0). No adverse events occurred, and she is undergoing ustekinumab

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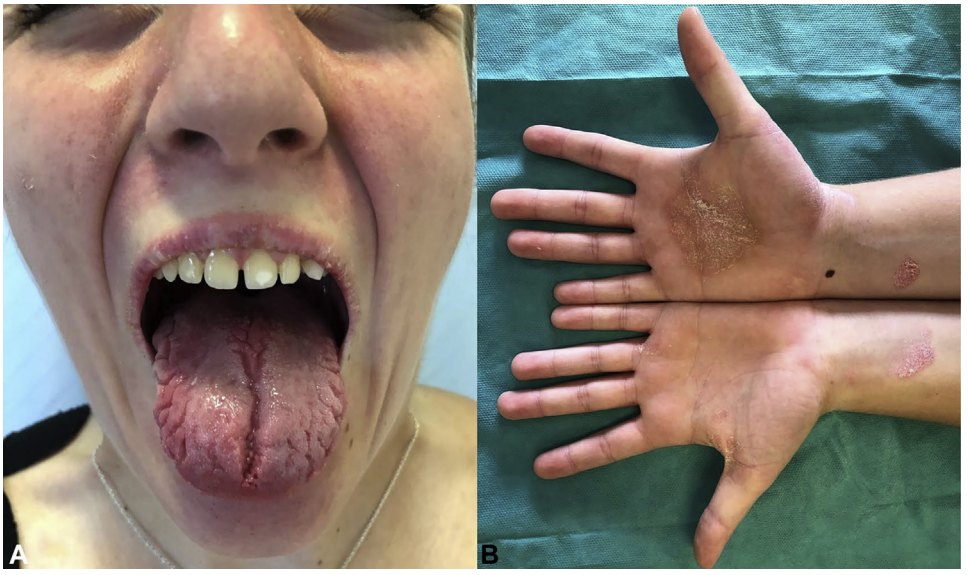

Fig 1. Fissured tongue (**A**) and palmar psoriasis (**B**) at presentation.

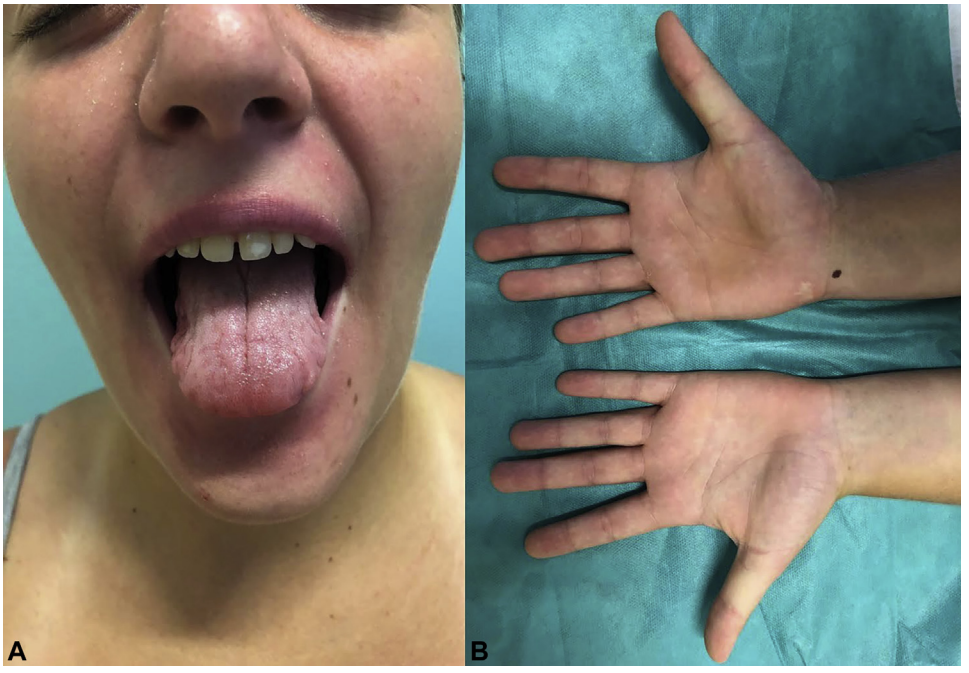

Fig 2. Resolution of tongue (**A**) and palmar psoriasis (**B**) after 16 weeks of treatment with 45 mg of ustekinumab.

treatment maintaining a PASI 0 response after 28 weeks of treatment.

DISCUSSION

Skin manifestations in sensitive areas are frequently observed, as facial involvement is reported in up to two-thirds of patients with psoriasis, whereas palmoplantar psoriasis and psoriatic glossitis is seen in 11% to 39% and 9.8% to 47.5% of patients, respectively.[3,4] Although geographic and fissured tongue are nonspecific oral manifestations that can be associated with psoriasis, a limited number of reports described a good response to antipsoriatic therapies (Table I).[5-8]

Psoriasis affecting sensitive sites may negatively affect QoL, with severe consequences on both daily and leisure activities, as in our patient. For this reason, these patients deserve appropriate

Table I. Previously described cases of psoriatic glossitis treated with antipsoriatic treatments

Study	Patient characteristics	Type of oral psoriasis	Associated form of psoriasis	Treatment	Treatment duration	Outcome
Younai and Phelan, 1997[5]	F, 65 y, white	FT	Plaque-type psoriasis of the lips	Erythromycin 250 mg 4-times daily, topical fluocinonide 0,05% ointment mixed with Orabase	5 d	Complete resolution
Casper et al, 1998[6]	F, 61 y, white	GT	generalized pustular psoriasis	acitretin, 35 mg/d	Unknown	Improvement of both oral and cutaneous lesions
	M, 84 y, white	GT + FT	generalized pustular psoriasis	acitretin, 70 mg/d + prednisolone	Unknown	Improvement of both oral and cutaneous lesions
Migliari et al, 2003[7]	M, 13 y, African	GT + FT	Plaque-type psoriasis of the lips	Topical retinoid and steroid association	Unknown	No clinical response
D'Erme et al, 2013[8]	M, 60 y, white	FT	severe plaque psoriasis (PASI, 16)	Infliximab (unknown dosing)	5 mo	Improvement of both oral and cutaneous lesions (PASI, 2)

FT, Fissured tongue; *GT*, geographic tongue.

treatment, even if their disease is limited in extension and cannot be considered severe (PASI<10). Based on national and international registry data, ustekinumab was proven the safest and the most effective therapy compared with other biologic agents.[9-12] Considering patients' preferences in the choice of treatment, a low treatment frequency was identified as a key factor for females, younger people, and working patients; therefore, it has been associated with an improved adherence.[13] Ustekinumab has the lowest number of injections per year, and its use was associated with an improvement in patients' adherence[13] compared with both tumor necrosis factor-α inhibitors and anti−interleukin-17 agents. The latter has yet to be approved in pediatric patients affected by psoriasis.

Among those agents approved to treat subjects younger than 18 years, 27 adalimumab injections occur during the first year of treatment (for children between 4 and 17 years of age and >30 kg, as in our case, injections must be administered every other week starting 1 week after a first dose of 40 mg), etanercept can be used as intermittent therapy, as a 24-week treatment (24 injections) and eventual retreatment after interruption can be prescribed in pediatric patients according to labeled instructions, whereas ustekinumab injections are 6 in the first year. To our knowledge, this is the first case of oral psoriasis successfully treated with ustekinumab, that induced a rapid remission (in 12 weeks) of psoriatic mucocutaneous manifestations and resolution of pain and discomfort associated with an improvement in patient's QoL.

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