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CASE REPORT

Lichenoid lesion on the tongue in a patient with Laugier-Hunziker Syndrome

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Abstract:

A 75-year-old woman presented with a painful lesion on the right lateral border of the tongue, seeking diagnosis and treatment. Clinical examination revealed erosions with whitish striae, consistent with a lichenoid lesion, which was confirmed by histopathology. The lesion was managed symptomatically with low-level laser therapy and monitored over time. In addition, the patient exhibited pigmented macules on the lips and longitudinal melanonychia. After ruling out other local and systemic conditions, a diagnosis of Laugier-Hunziker syndrome (LHS) was established. Laugier-Hunziker syndrome (LHS) is a rare, benign pigmentary disorder characterized by slate-gray to dark brown macules on the mucosa, nails, and acral sites. It predominantly affects individuals of French and Italian descent, but it has also been observed in Hispanics, Arabs, and Asians. To date, approximately 200 cases of LHS have been described in the literature, with no documented association with oral lichenoid lesions. This report illustrates the concomitant occurrence of these two conditions, an association not yet recognized in the literature, which should be considered anecdotal so far. This report highlights the importance of thorough clinical evaluation and awareness of rare conditions, such as LHS, to prevent misdiagnoses and enhance understanding of its potential associated manifestations.

Keywords: Laugier-Hunziker syndrome; Oral pigmentation; Melanonychia; Lichenoid lesion; Case report.

INTRODUCTION

Laugier-Hunziker syndrome (LHS), also known as Laugier-Gerbig-Hunziker syndrome, Laugier-Hunziker-Baran syndrome, or idiopathic lenticular mucocutaneous pigmentation, is an uncommon benign condition characterized by hyperpigmented macules affecting the oral mucosa, nails (melanonychia), and acral skin areas[1]. This syndrome was first described in 1970, by Laugier and Hunziker[2].

LHS primarily manifests in middle-aged adults, with a mean age of onset around 50 years, and is more common after puberty. The condition shows a higher prevalence in women and has been most frequently described in white individuals, especially of French and Italian descent[3].

Although its etiology is not yet fully understood, LHS has occasionally been associated with genetically based disorders in some literature reports[4].

The proposed pathogenesis involves melanocytic dysfunction, resulting in increased synthesis of melanosomes and their transfer to the basal epithelial layers[3].

LHS is a benign condition, with no associated systemic manifestations, of insidious onset and chronic course, with no known risk of malignant transformation. The absence of constitutional symptoms (fatigue, weight loss), dysmorphic features, and signs of gastrointestinal or cardiovascular compromise — combined with normal exams and labs — supports the diagnosis by exclusion[5].

Statement of Clinical Significance
This case highlights the importance of differential diagnosis in oral pigmentation, contributing to the identification of Laugier-Hunziker Syndrome and its novel association with lichenoid lesion, guiding more accurate and safer clinical approaches.

Despite being harmless, LHS can mimic serious systemic diseases such as Peutz-Jeghers syndrome, Addison's disease, and McCune-Albright syndrome[6-8].

Some case reports describe associations between LHS and conditions like esophageal melanocytosis, actinic lichen planus, bone marrow hypocellularity, thrombocytopenia, invasive melanoma, and lupus erythematosus[9].

LHS has also been found in patients with autoimmune disorders such as systemic lupus erythematosus,

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rheumatoid arthritis, Sjögren's syndrome, autoimmune hemolytic anemia, and bullous pemphigoid[10].

The term "oral lichenoid lesion" (OLL) was introduced by Finne in 1982 to describe oral changes associated with medications, restorative materials, flavorings, allergies, and systemic diseases, such as autoimmune hepatitis[11].

OLLs present as reticulated, papular, or plaque-like whitish lesions, often accompanied by erythema or erosion. They may be asymptomatic or cause burning or pain[12]. The distinction of OLLs and oral lichen planus (OLP) is based on clinical and histopathological criteria (Table 1)[13].

To our knowledge, there are no previous reports of LHS occurring together with lichenoid lesions.

CASE REPORT

A 75-year-old white woman was referred to the Oral Diagnosis Clinic (FOUSP) with a complaint of pain in a lesion on the right lateral tongue present for three months. She also reported a similar lesion on the left side.

Her medical history included systemic arterial hypertension and an acute myocardial infarction 13 years previously. The patient was continuously taking atenolol, losartan, levothyroxine, aspirin, and atorvastatin — indicating polypharmacy. However, clinical evaluation did not identify or report hyposalivation.

The patient was undergoing regular medical follow-up and had recent routine exams without significant abnormalities, with no clinical evidence of gastrointestinal disorders, nutritional deficiencies, or associated systemic conditions.

Furthermore, the patient had been wearing the same upper complete denture and lower removable partial denture for approximately 15 years. During the clinical examination, it was observed that the dentures were unsatisfactory in terms of hygiene, which could favor fungal colonization and contribute to the symptoms. Therefore, topical antifungal therapy was included in the treatment plan. The patient reported being a former smoker, having stopped smoking 35 years ago.

Intraoral examination revealed irregular erosions with whitish streaks interspersed with erythematous areas on the right lateral border of the tongue, as well as mild erythema with white streaks on the left lateral border. Discrete, confluent brownish macules were symmetrically distributed on the upper and lower labial mucosa (Figure 1).

The initial diagnostic hypothesis included oral lichenoid lesion, oral lichen planus, adverse drug reaction, and erythroplakia.

As part of the initial evaluation, a tissue fluorescence examination was performed with the VELscope device, a non-invasive auxiliary method that detects changes in autofluorescence of the oral mucosa and aids in the early identification of areas suspected of dysplasia or malignancy. The examination revealed loss of autofluorescence in the lesioned areas, which reinforced the indication for an incisional biopsy to investigate possible dysplasia or malignant transformation (Figure 2).

Histopathological examination revealed parakeratinized stratified squamous epithelium exhibiting mild exostosis and acanthosis. The underlying connective tissue is dense and presents focal areas with a discrete mononuclear inflammatory infiltrate close to

Table 1. Histopathological features of oral lichen planus and oral lichenoid lesions[13].

Histopathological features	OLP	OLL
Surface keratinization	Hyperkeratosis (ortho or para)	Often present, sometimes more pronounced
Epithelial architecture	Irregular acanthosis, "saw tooth" rete pegs	Atrophy or spongiosis
Basal cell layer	Hydropic degeneration Civatte bodies	Similar features
Inflammatory infiltrate	Band-like lymphocytic infiltrate in superficial lamina propria	Mixed inflammatory infiltrate, plasma cells and eosinophils
Inflammation distribution	Subepithelial	Perivascular and deeper in the connective tissue
Vascular changes	Not prominent	Perivascular inflammation
Epithelial dysplasia	Absent	May be present
Clinical	Bilateral and symmetrical	Often unilateral or non symmetrical

OLP: oral lichen planus; OLL: oral lichenoid lesions.

the epithelium. Striated muscle bundles were observed beneath the epithelium in the deeper connective tissue. No dysplastic changes were observed (Figure 3).

The patient underwent treatment with oral miconazole (20 mg/g gel, applied four times daily, one week) and weekly photodynamic therapy (PDT) sessions, totaling three consecutive sessions. There was complete resolution of symptoms at the end of treatment.

During clinical evaluation, long-standing brownish pigmentation of the toenails was also reported, which the patient had previously interpreted as a fungal infection. The association of pigmented labial macules with melanonychia striata and the absence of gastrointestinal disorders or family history led to the diagnosis of Laugier-Hunziker syndrome (LHS) concomitant with oral lichenoid lesion (Figure 4A-D).

DISCUSSION

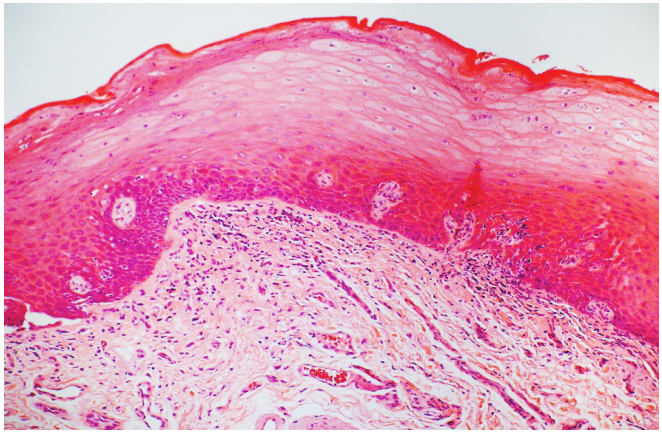

Figure 3. Histopathological section showing parakeratinized stratified squamous epithelium with mild exostosis and acanthosis. The underlying connective tissue is dense, with focal areas of discrete mononuclear inflammatory infiltrate adjacent to the epithelium. Striated muscle bundles are present in deeper layers. No evidence of epithelial dysplasia was found.

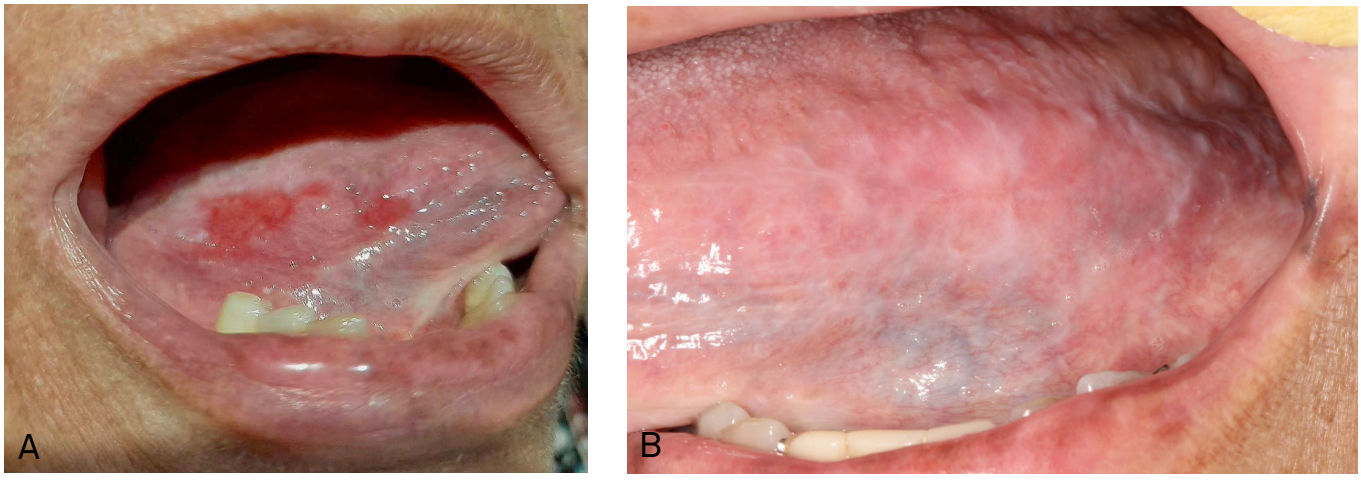

Figure 1. (A) Right lateral border of the tongue showing erythema interspersed with delicate white striae. **(B)** Left lateral border showing whitish striae without associated erythema.

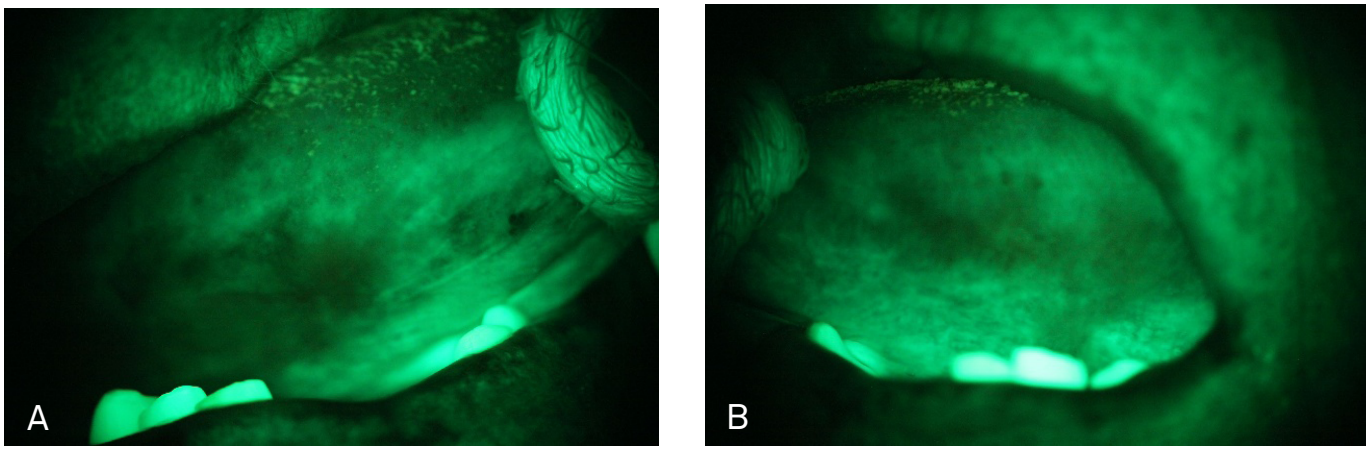

Figure 2. Autofluorescence examination using the VELscope® device. **(A)** Right lateral border of the tongue showing loss of fluorescence in the lesioned area. **(B)** Left lateral border with mild fluorescence alteration in line with the clinical aspect observed.

Laugier-Hunziker syndrome (LHS) is characterized by lentiginous hyperpigmentation of the oral mucosa and lips. In addition, longitudinal melanonychia and hyperpigmented palmar and plantar lesions may occur[14].

Identifying LHS helps exclude other more serious syndromes associated with hyperpigmentation[15] (Table 2).

The main entities included in the differential diagnosis due to similar clinical presentation are Addison's disease and Peutz-Jeghers syndrome (PJS), both of which are characterized by multiple macules on the oral mucosa. In Addison's disease, patients also present systemic signs such as hypotension, hyponatremia, hyperkalemia, hypoglycemia, and elevated blood urea nitrogen, resulting from adrenal insufficiency. PJS is an autosomal dominant hereditary condition that presents with mucocutaneous pigmentation associated with gastrointestinal polyposis and, most importantly, an increased risk of various types of neoplasms[16].

Another endocrine condition that may present with cutaneous hyperpigmentation is McCune-Albright syndrome. However, it usually manifests early in life and is characterized by fibrous dysplasia, café-au-lait macules, and endocrine hyperfunction, with segmental-pattern skin lesions[17]. Pigmentation induced by medications varies depending on the agent involved, presenting them with different patterns and shades. Drugs such as antipsychotics, anticonvulsants, antimalarials, cytotoxics, amiodarone, and NSAIDs are among the main causes[18]. Despite the polypharmacy that characterized our patient, there were no drugs typically associated with pigmentation that more often affects the palate, gingiva, or buccal mucosa[19].

The finding of asymptomatic mucosal pigmentation associated with longitudinal melanonychia in the absence of systemic manifestations supported the diagnosis of Laugier-Hunziker syndrome (LHS). However, anecdotal reports in the literature describe the occurrence

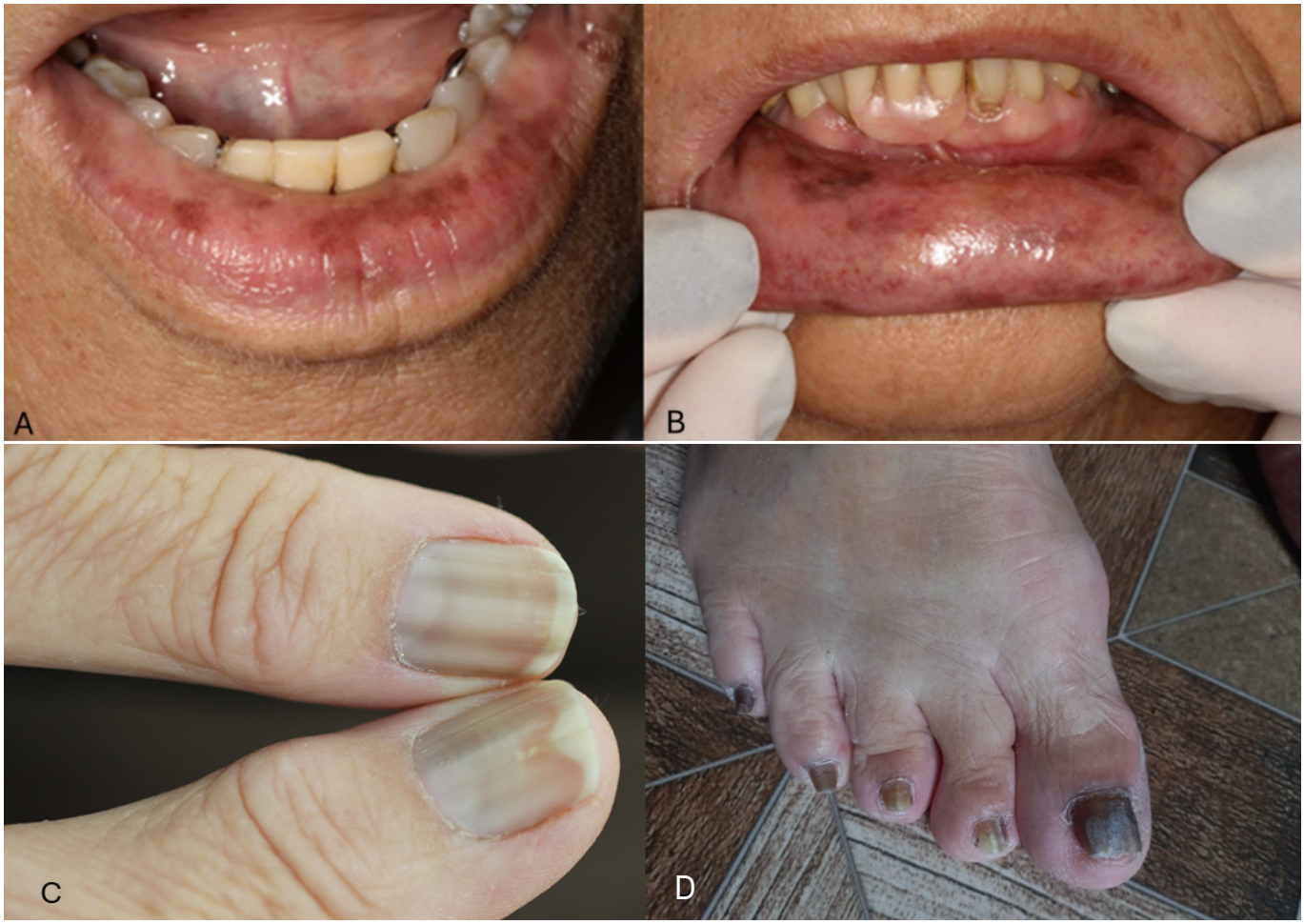

Figure 4. Pigmentations characteristic of Laugier-Hunziker syndrome: dark brown melanotic macules on the lips **(A, B)**, associated with longitudinal melanonychia on the fingernails **(C)** and toenails **(D)**.

of LHS in patients with autoimmune diseases such as Sjögren's syndrome, systemic lupus erythematosus, autoimmune hemolytic anemia, and inflammatory arthritis[20]. This prompted a re-evaluation of the lichenoid lesions and their potential etiological associations.

Lichenoid lesions exhibit a histopathologic pattern characterized by dense lymphocytic infiltrate at the epithelium-lamina propria interface, vacuolar degeneration of basal cells, and keratinocyte apoptosis, reflecting a chronic inflammatory process typical of conditions such as oral lichen planus, which is clinically characterized by symmetrical lesions always involving buccal mucosa[11,21,22]. Clinically, these lesions present as erythematous, white, or reticulated plaques that may cause symptoms such as burning or discomfort. The etiology of lichenoid lesions is multifactorial, involving drug reactions, exposure to irritants, and immunological processes[23].

There is no curative option for oral lichen planus (OLP) or oral lichenoid lesions (OLL) due to the lack of a clear etiology. The therapies available are essentially symptomatic. Currently, topical corticosteroids are the most widely used treatment for oral lichen planus (OLP), along with other immunomodulatory agents, retinoids, ultraviolet irradiation, and/or laser therapy[11]. Corticosteroids reduce inflammation and symptoms of lichenoid lesions by suppressing the immune response. They alleviate pain and burning but may cause local adverse effects such as candidiasis and recurrences after withdrawal[21]. Comparative studies indicate that photodynamic therapy (PDT) is similarly effective as topical corticosteroids, but with fewer side effects[24].

PDT has emerged as a promising approach in the treatment of OLP, especially in symptomatic cases or those resistant to conventional corticosteroid therapy. In PDT, a photosensitizing agent is activated by a specific light source, generating free radicals that induce selective apoptosis in inflammatory and altered cells, with immunomodulatory and antimicrobial effects and minimal damage to healthy tissues. Studies indicate that this therapy can significantly reduce lesion size and associated symptoms, such as pain and discomfort, with a low rate of side effects. Thus, PDT represents an effective, safe, and minimally invasive therapeutic alternative for managing oral lichenoid lesions[25].

In the reported case, dark brown to blackish-gray macules were observed on the lips, with irregular

Table 2. Comparative clinical features among Laugier-Hunziker Syndrome, Peutz-Jeghers Syndrome, Addison's Disease, and McCune-Albright Syndrome.

	LHS	PJS (Peutz-Jeghers)	Addison's Disease	McCune-Albright
Etiology	Idiopathic	Genetic (STK11 mutation)	Endocrine (adrenal insufficiency)	Post-zygotic GNAS mutation
Clinical appearance	Lenticular pigmented macules	Perioral and mucosal pigmented macules	Diffuse mucosal and skin hyperpigmentation	Café-au-lait macules
Color	Brown to dark gray	Dark brown	Dark brown	Light brown
Distribution	Lips, buccal mucosa, tongue	Lips, oral mucosa, perioral region, fingers	Oral mucosa, gingiva, skin	Trunk, face, occasional mucosa
Symptoms	Asymptomatic	Asymptomatic (possible intestinal symptoms)	Systemic symptoms: fatigue, hypotension	Early endocrine dysfunctions
Clinical course	Stable, benign	Chronic, hereditary	Progressive if untreated	Chronic with variable manifestations
Systemic association	None	Intestinal polyps, cancer risk	Adrenal insufficiency	Hormonal dysfunctions, fibrous dysplasia
Nail findings	Longitudinal melanonychia	Rare	Possible	Uncommon
Malignancy potential	No	Yes (gastrointestinal)	Not directly, but related to endocrine causes	Yes (osteosarcoma, breast cancer)
Histopathology	Increased melanin without melanocytic proliferation	Basal melanosis	Increased melanin in basal layer	Epidermal melanosis
Management	Follow-up	Follow-up, cancer screening	Hormone replacement	Endocrine and bone treatment

LHS: Laugier-Hunziker Syndrome; PJS: Peutz-Jeghers Syndrome.

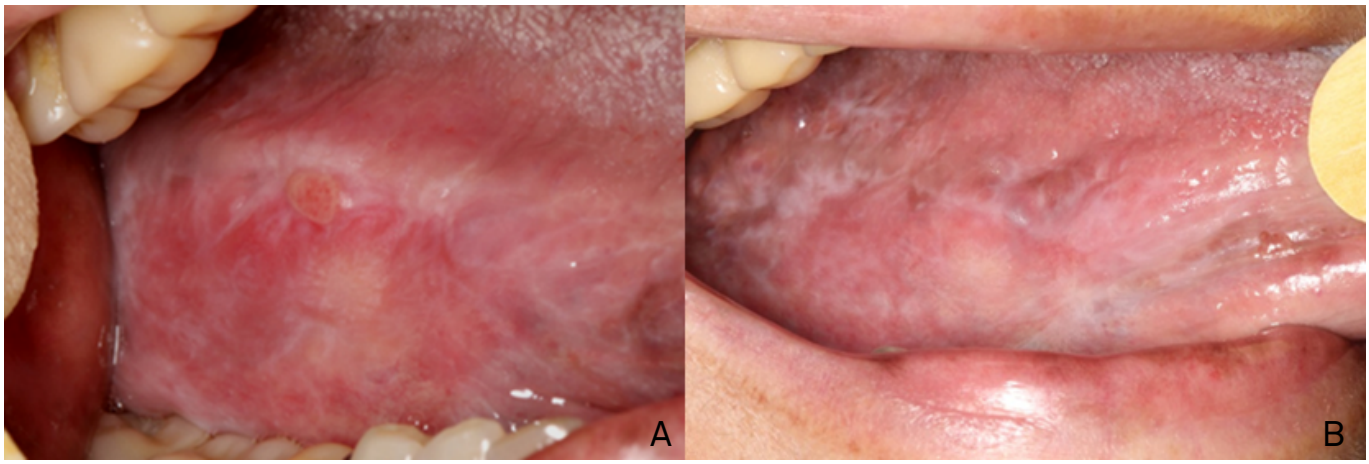

Figure 5. Clinical aspect of the oral lichenoid lesion with characteristic whitish streaks, **(A)** highlighting the region subjected to incisional biopsy. **(B)** Image after treatment shows a reduction in erythematous areas and a decrease in inflammatory characteristics.

contours and variable size. Pigmented striations were present on multiple fingernails and toenails, with no evidence of dysplasia or malignancy.

The main complaint — symptomatic lichenoid lesion — was successfully treated with PDT, emphasizing the importance of considering concomitant pathologies. The patient was included in a regular follow-up regimen to monitor the lichenoid lesion. This case is also notable for the misdiagnosis of nail pigmentation as a fungal infection, revealing limited knowledge about LHS among healthcare professionals (Figure 5).

CONCLUSION

The association between Laugier-Hunziker syndrome (LHS) and oral lichenoid lesion (OLL), as observed in this case, is unprecedented in the literature and highlights the importance of a comprehensive clinical examination of the patient, regardless of the patient's chief complaint, especially in atypical or overlapping clinical presentations. The absence of systemic manifestations and relevant family history supported the diagnosis of LHS. Simultaneously, the presence of symptomatic lesions compatible with OLL required a specific therapeutic approach, successfully achieved using photodynamic therapy, complemented by topical antifungal treatment due to prosthetic conditions. This report emphasizes the importance of differential diagnosis and multidimensional assessment, suggesting that the coexistence of LHS and OLL, although rare, may occur and require distinct yet complementary clinical approaches. Additionally, it reinforces the need for greater awareness among healthcare professionals regarding LHS and its potential associations with other mucosal conditions.

AUTHORS' CONTRIBUTIONS

FSO: Conceptualization, Investigation, Writing – original draft. MAT: Investigation, Validation, Visualization. CAL: Project administration, Supervision, Validation, Writing – review & editing. FDN: Investigation, Formal analysis. NNS: Project administration, Supervision, Validation, Writing – review & editing.

CONFLICT OF INTEREST STATEMENT

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