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RESEARCH ARTICLE

PALMOPLANTAR LICHEN SCLEROSUS ET ATROPHICUS : A CASE REPORT

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Abstract

Introduction: Lichen sclerosus et atrophicus mainly affects the genital area. The palmo-plantar involvement is most often part of a diffuse cutaneous involvement, which is rarely exclusive. We report a case of acral lichen sclerosus et atrophicus, without any other involvement.

Case Report: A 58-year-old man presented with lesions of the hands and feet, consisting of subungual hyperkeratosis and squamo-keratoticperi-onyxis of the toenails, sclero-atrophic ivorine plaques with depressed keratoticmicropapules on the soles and palm hollows, extending to the lateral and dorsal surfaces, with involvement of the interdigital and inter-toe spaces. There was no other cutaneous or genital involvement. Histological examination showed a lichen sclerosus et atrophicus appearance.

Discussion: A few cases of lichen sclerosus et atrophicus limited exclusively to the feet and/or hands have been reported, affecting either the palmoplantar or dorsal surface, with or without ungual involvement. No case of lichen sclerosus et atrophicus affecting only the feet and hands on both surfaces with onychodystrophy, as in our case, has been reported.

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Introduction:-

Lichen sclerosus et atrophicus (LSA) is a chronic inflammatory dermatosis, and predominantly female. It mainly affects the genital area, cutaneous involvement is uncommon, while palmo-plantar localization remains rare.

We report a rare case of acral LSA, affecting the palms and soles with extension to the dorsal surfaces.

Case Report:

Patientis58 yearsold, chronic smoker. He consulted for palmo-plantar papular lesions, painful when walking, evolving for 12 years. The dermatological examination showed hypopigmented, ivory-colored, finely scaly lesions poorly limited, associated with depressed keratoticmicropapules on the soles of the feet, extending to the lateral and dorsal surfaces, with a scleroatrophiccicatricial evolution. Onychodystrophic toenails with subungual hyperkeratosis and squamo-keratoticperi-onyxis. There were rounded to oval plaques, confluent in scleroatrophic ivory placards with jagged edges, on the thenar and hypothenar eminences and on the palmar surface of the fingers, extending to the lateral surfaces and spreading in small lesions on the anterior surface of the wrist. There were small depressed keratotic papules in the hollows of the palms (Figure 1). The interdigital and inter-toe spaces were also affected.

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These lesions were painful to palpation. The rest of the skin integument and the genital mucosa were intact. The rest of the somatic examination was unremarkable.

The histological study was in favor of lichen sclerosus et atrophicus, showing an atrophic epidermal coating associated with an orthokeratotic hyperkeratosis and whose basal membrane is the seat of a degeneration of lichenification associated with lymphocytic exocytosis, realizing a mottled appearance of the basal layers; the superficial dermis and the middle dermis are poorly represented and are the site of a dense inflammatory infiltrate in a band parallel to the mucous body, composed of lymphocytes (figure 2). The serologies of human immunodeficiency virus (HIV) and viral hepatitis B and C were negative. A thoracoabdomino-pelvic CT scan was performed as part of the paraneoplastic workup and showed no abnormalities.

The patient was initially put on a treatment based on topical keratolytics and dermocorticoids for3 months, he did not show any improvement which led to the change of the therapeutic line by methotrexate with a failure after 6 months. The patient was put on acitretin. After 6 months of acitretin and topical treatment, pain subjectively improved and acral hyperkeratosis decreased, resulting in improved patient comfort and quality of life. Atrophic plaques and erythema on the palmoplantar surfaces and overlying dorsal areas remained.

Discussion:-

Lichen sclerosus et atrophicus (LSA) is a chronic inflammatory dermatosis of unknown etiology, but it is associated with several factors: autoimmunedisorders, genetic (HLA class II DQ7), infectious (human papillomavirus, Borreliaburgdorferi) and traumatic by Köebner's phenomenon [1,2,3]. It affects subjects of all ages with a female predominance [1,2]. Lichen sclerosus mainly affects the ano-genital region, whereas skin involvement is seen in only 20% of cases, half of which are associated with genital involvement [3,4]. Purely extragenital localization occurs in 10% of patients [2,3].

Acral involvement in lichen sclerosus has been described in a few cases in the literature. Most of them were part of more diffuse skin involvement, affecting the limbs or the trunk, or were associated with anogenital lesions [4]. Exclusive acral localization seems to be exceptional [3,4].

The first reported patient with palmar involvement of LSA was described by Purres and Krull [5] in 1971; the palmar location was associated with extensive skin involvement. While the first case of plantar LSA without other skin manifestations was described in 1978 by Hammar [6]. In 1979, Petrozzi et al [7] reported a case of LSA limited exclusively to the palms and soles. Since then, there have been only a few reports of exclusive acralinvolvement.The reported cases of palmoplantar involvement during LSA are summarized in Table I.

In general, extragenital LSA is asymptomatic, sometimes affected patients may experience pruritus or irritation[2,3]. Many observations reported that the acral involvement was painful, causing functional disability and altering the patient's quality of life [2,3,4,11].

LSA presents as pearly white papules, which may coalesce into plaques with crumbled edges, more or less atrophic, sometimes surrounded by a peripheral erythematous halo or centered by a corneal protrusion.

At the acral level, it presents as ivory yellow keratotic papules that coalesce into plaques or as atrophic erythematous patches [2,4] with accentuated eccrineostia or prominent dermatoglyphs [2], and sometimes as telangiectasias, purpura, hemorrhagic blisters, and fissures [3,11]; bullous variants have been reported in the literature [2,8]. Thus, an involvement of the dorsal surfaces has also been described [4,12,13,14] and of the interdigital spaces [3,4] with sometimes an onychopathy [4,12,14] in the form of onychodystrophy or striated nails.

The histological appearance of early LSA lesions includes banded lymphocytic inflammatory infiltrate in the superficial dermis and vacuolar degeneration of the basal layer. In older lesions, orthohyperkeratosis, epidermal atrophy, vacuolation of the basal layer, homogenization of the superficial dermis, with hyalinization and sclerosis of the papillary dermis, dermal edema, and disappearance of elastic fibers are observed [2,4,11]. Histologically, the main differential diagnosis is lichen planus in early lesions, whereas the histological appearance of very old lesions may resemble morphea [4,11].

First-line treatment of LSA includes potent topical steroids. Other alternatives: topical testosterone or progesterone ointment, topical or systemic retinoids, systemic immunomodulators, topical calcineurin inhibitors, phototherapy and photodynamic therapy. Surgical interventions have also been reported as therapeutic options in case of complications. [2,3,11] Oral antimicrobials have been proposed by some authors because of the possibility of association with Borrelia infection. [15] Hydroxychloroquine (400 mg/d) showed improvement in one case of plantar LSA. [3]

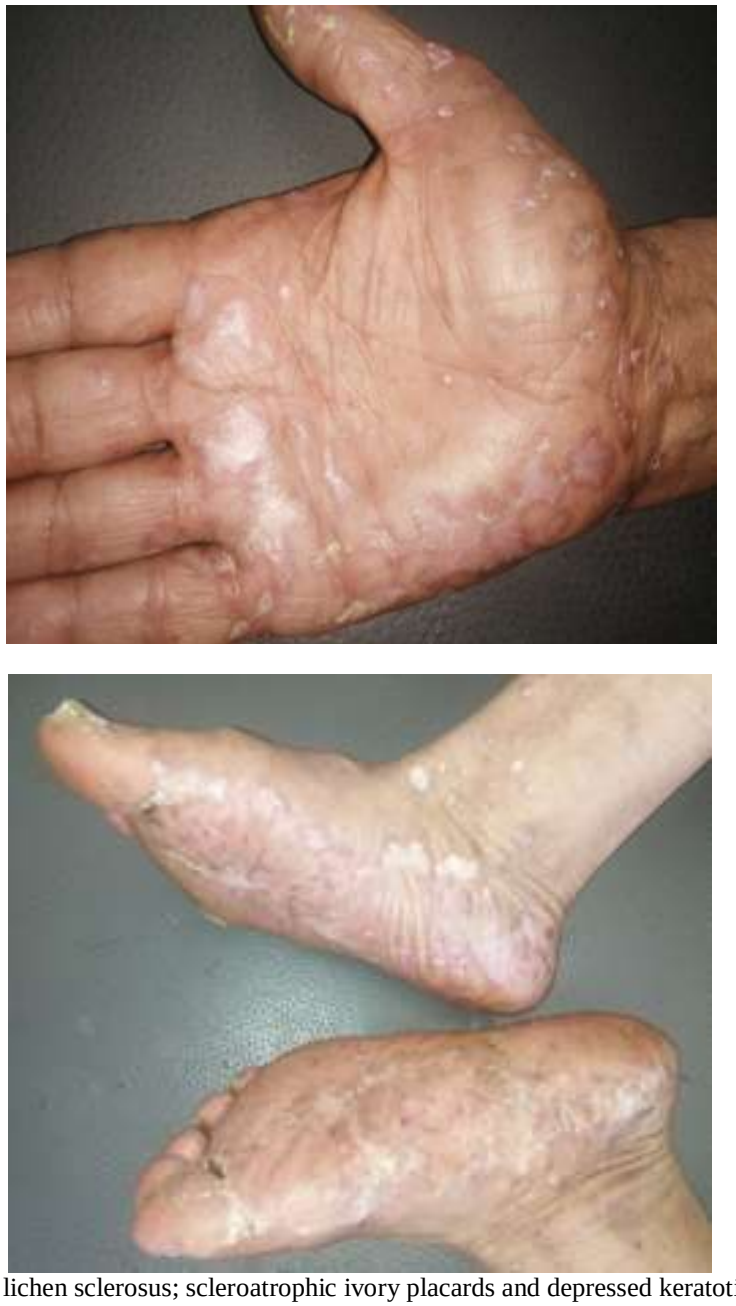

Figure 1:- Palmoplantar lichen sclerosus; scleroatrophic ivory placards and depressed keratotic micro-papules.

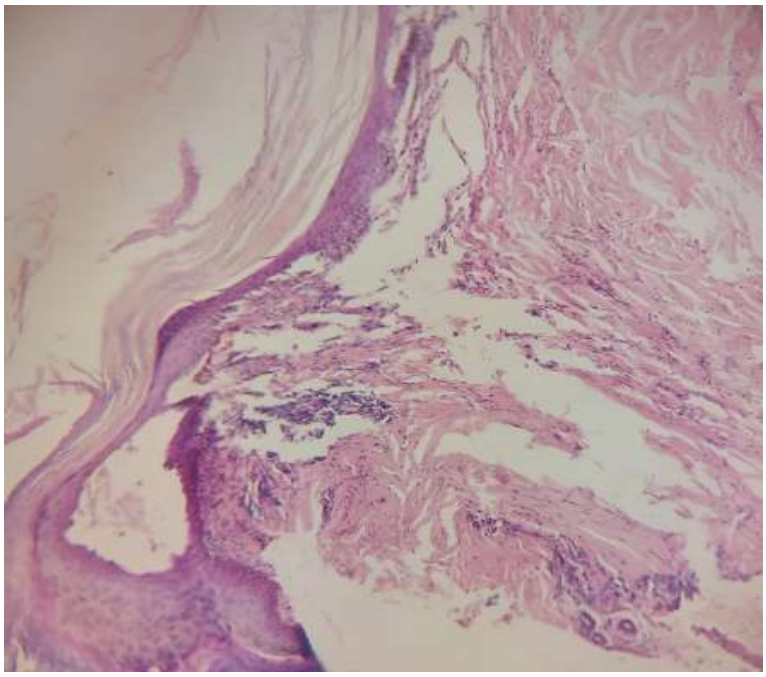

Figure 2:- Histological image of lichen sclerosus.

	Purres and Krull [5]	Hammar [6]	Petrozzi and al [7]	Tudino and Wong [8]	Aggarwal and al [9]	Steff and al [4]	Caspary and al [3]	Viana and al [10]	Gómez-Martín and al [11]	Seyffert and al [2]	El Bakali and al
Sex	Female	Male	Female	Female	Male	Female	Female	Female	Female	Female	Male
Age, y	71	N/A	57	54	58	56	63	57	78	69	58
Comorbid conditions	Healthy	Erythro-melalgia	Healthy	N/A	N/A	-	DM 2 HTG	-	HTN, DM 2 Dyslipidaemia minor depression	Smoker, HTN, DM2 Hypothyroidism COPD Hyperlipidemia	Smoker
Genital involvement	+	-	-	-	-	+	-	-	+	-	-
Palmar involvement	+	-	+	+	+	+	+	+	-	+	+
Plantar involvement	-	+	+	-	+	+	+	+	+	-	+
Onychopathy	-	-	-	-	-	+	-	-	-	-	+
dorsal face involvement	-	-	-	-	-	+	-	-	-	-	+
Generalized cutaneous involvement	+	-	-	+	+	+	-	+	-	+	-
Treatment	Topical and intralesional Steroids	N/A	Keratolytics, Topical steroids	Topical steroids	N/A	Topical steroids	Topical steroids, oral HCQ	calcipotriol, topical calcineurin inhibitors, Topical steroids	Keratolytics, Topical steroids	Topical steroids, topical calcineurin inhibitors, methotrexate, isotretinoin	Keratolytics, Topical steroids, methotrexate, isotretinoin
Treatment response	Refractory	N/A	Refractory	Partial success	N/A	N/A	improvement	improvement	improvement	Partial success	Partial success

Table I: Comparison of reported cases of palmar and / or plantar lichen sclerosus

Conclusion:-

LSA, with exclusive acral localization, is rare. It should be considered as a differential diagnosis of whitish and atrophic palmoplantar lesions. Concerning our case of palmoplantar LSA with extension to the dorsal surfaces, and involvement of the nails and inter-digital spaces has never been reported in the literature.

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