Giant Squamous Cell Carcinoma on Chronic Lichen Planus on the Ankle - A Case Report and Short Literature Review

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Abstract

BACKGROUND: Cutaneous squamous cell carcinoma (SCC) is the second most common malignancy of skin. Although a major risk factor is a chronic exposure to ultraviolet radiation, preexistant chronic inflammatory disorders may also possess an increased risk for SCC. That is not the case for cutaneous lichen planus in contrast to oral lichen planus and oral SCC.

CASE REPORT: We report the case of an 87-year-old Caucasian woman presenting with a giant verrucous tumour on the left ankle. She suffered from long-standing disseminated lichen planus. Histology confirmed the diagnosis of SCC on partly verrucous lichen planus. The course was complicated due to sepsis. An emergency transfemoral amputation became necessary. The patients survived and could be released into her nursery. A literature review underlined the rarity of SCC on lichen planus of the skin. Most of these rare cases were in patients in their second half of life on the lower legs. Hypertrophic lichen planus was overrepresented.

CONCLUSIONS: Although very rare by number, SCC can complicate lichen planus and lead to the life-threatening situation. Atypical verrucous lesions on lichen planus warrant a histologic analysis. Surgery is the treatment of choice for cutaneous SCC.

Introduction

Cutaneous squamous cell carcinoma is the second most common malignancy of skin. Chronic exposure to ultraviolet radiation (UVR) is the most important environmental factor. Fair skin complexion, immunosuppression, exposure to arsenic compounds are also contributing factors [1].

Major risk factors for relapse and metastasis are a Breslow thickness > 6 mm (Relative risk-RR, 7.13) and perineural invasion (RR, 4.30), while a diameter > 20 mm (RR, 3.22) bears the highest risk for metastasis [2]. SCC can develop as a consequence of chronic inflammatory skin lesions such as in chronic discoid lupus erythematosus [3], hidradenitis suppurativa/ acne inversa [4], chronic leg ulcers [5], erythema ab igne [6], or recessive dystrophic epidermolysis bullosa [7], and lichen planus [8].

Lichen planus is a chronic inflammatory T-cell disorder of the skin and mucous membranes. Oral lichen planus is considered a facultative precancerous lesion for oral SCC. Ulcerations, tongue site, and female gender are risk factors for a malignant transformation of oral lichen planus into SCC. The overall transformation rate has been estimated in one study as high as 1.4% [8]. On the contrary, a meta-analysis for cutaneous lichen planus came to the conclusion that there is no increased risk for cutaneous SCC [9].

Case Report

An 87-year-old female patient from a nursery presented with a malodorous verrucous tumour on her left leg. She had a medical history of a complicated...
lower leg fracture 30 years ago. She had no weight loss or fever but reported a local pain.

![Figure 1: Disseminated lichen planus of the leg (right leg)](image)

On examination, we observed a circumferential exophytic verrucous tumour on a chronic lichen planus lesion on the left ankle (Figure 1 and 2).

![Figure 2: Circumferential verrucous squamous cell carcinoma on lichen planus (left leg)](image)

We performed a deep skin biopsy for diagnosis. Histologic examination revealed a well-differentiated cutaneous squamous cell carcinoma (SCC), partly ulcerated, with bacterial colonization (Figure 3) on a verrucous lichen planus on the left leg and partly verrucous lichen planus on the right leg and left lower arm (Figure 4).

![Figure 3: Histology of the squamous cell carcinoma composed of islands and cords of epithelial cells with keratin pearls in deep dermis (HE x 4)](image)

Lymph node ultrasound demonstrated several suspicious enlarged nodes in the left groin with up to 42 mm in diameter. X-ray of the left lower suggested old posttraumatic lesions with inhomogenous spongiosa of the distal third of the tibia bone. Chronic osteomyelitis could be ruled out for sure. A bony infiltration of the tumour was possible.

Computerised tomography of the trunk excluded a metastatic spread. Enlarged lymph nodes in the groins were considered to be reactive only.

![Figure 4: Histology of lichen planus with sawtooth-like epidermis, hyperkeratosis and hypergranulosis associated](image)

Laboratory findings: Leucocytosis of 24 Gpt/L (normal range 4 to 10), stable hypochromic microcytic anaemia of with a haemoglobin level of 6 mmol/L (normal range 8.4 to 10.9), lactate dehydrogenase was normal.

The final diagnosis was SCC (pT3 cNX cM0 G1) on verrucous lichen planus.

The case was brought to the interdisciplinary tumour board. Radiotherapy was not considered to be curative, and chemotherapy was hardly tolerable by the elderly patient. Vascular and orthopaedic surgeons suggested a major amputation of the left leg, but the patient disagreed. We performed palliative therapy and good ulcer care.

Two weeks later, she developed fever, hypotonia, tachycardia and fatigue. Her procalcitonin level was 37 ng/mL. A venous blood culture identified the anaerobic germ *Bacteroides fragilis*. Laboratory findings disclosed leukocytosis of 24.7 Gpt/L, anaemia 5.30 mmol/L, thrombocytosis of 517 Tpt/L, and a C-reactive protein of 60.9 mg/L.

She was treated in the emergency department for sepsis. Three days later, a transfemoral amputation was necessary. Histological analysis revealed a verrucous SCC, R0 resection, and a verrucous lichen planus. After surgery, the patient rejected lymph node removal, radio or chemotherapy. Final tumour diagnosis was verrucous SCC pT3 NX G1 R0. The patient could be released into her nursery.
Discussion

In contrast to oral lichen planus, development of an SCC on cutaneous lichen planus is rare. The incidence of SCC in lichen planus has been estimated between 0.4% [9] and 1.74% [10].

In the meta-analysis of Sigurgeirsson and Lindelöf (1991) 36 reports of SCC associated with lichen planus were analysed amongst 2071 lichen planus patients [10]. In 2011, Friedl et al. reported on three patients who developed an SCC on chronic lichen planus of the lower leg and analysed another 24 cases within five years [11]. Five more cases have been reported since then in the international literature, mostly on lower legs and in association with hypertrophic or verrucous lichen planus [12], [13], [14], [15], [16]. We identified another two cases between published 2003 and 2007 [17], [18]. This translates into only 72 cases, including the present one.

During the process of malignant transformation, keratinocytes lose the production of C-Jun, part of the AP-1 transcription factor complex for the regulation of cell proliferation, differentiation and transformation [19]. The late epidermal differentiation marker K2e, a polypeptide of 70 kd, becomes diminished in cutaneous SCC [20].

Cutaneous lichen planus, however, does not possess an increased risk for SCC [10]. These cases of concomitant lichen planus and SCC are extremely rare.

References

1. Green AC, Olsen CM. Cutaneous squamous cell carcinoma: an epidemiological review. Br J Dermatol. 2017; 177(2):373-381. https://doi.org/10.1111/bjd.15324 PMid:28211039
2. Thompson AK, Kelley BF, Prokop LJ, Murad MH, Baum CL. Risk Factors for Cutaneous Squamous Cell Carcinoma Recurrence, Metastasis, and Disease-Specific Death: A Systematic Review and Meta-analysis. JAMA Dermatol. 2016; 152(4):419-28. https://doi.org/10.1001/jamadermatol.2015.4984 PMid:26762219 PMCID:PMC4833641
3. Motswaledi MH, Khammissa RA, Wood NH, Meyerov R, Lemmer J, Feller L. Discoid lupus erythematosus as it relates to cutaneous squamous cell carcinoma and to photosensitivity. SADJ. 2011; 66(7):340-3.
4. Chapman S, Delgadillo D III, Barber C, Khachemoune A. Cutaneous squamous cell carcinoma complicating hiradenitis suppurativa: a review of the prevalence, pathogenesis, and treatment of this dreaded complication. Acta Dermatovenerol Alp Pannonica Adriat. 2018; 27(1):25-28. https://doi.org/10.15570/actaapa.2018.5 PMid:29589641
5. Reich-Schupke S, Doerfer M, Wolinia U, Dissemond J, Horn T, Strölin A, Ersfurth-Berge C, Stücker M. Squamous cell carcinomas in chronic venous leg ulcers. Data of the German Marjolin Registry and review. J Dtisch Dermatol Ges. 2015; 13(10):1006-13. https://doi.org/10.1111/ddg.12649
6. Wolinia U, Helm C, Hansel G, Köstler E, Schönglebe J. Two cases of erythema ab igne, one with a squamous cell carcinoma. G Ital Dermatol Venereol. 2007; 142(4):415-418.
7. Kim M, Murrell DF. Update on the pathogenesis of squamous cell carcinoma development in recessive dystrophic epidermolysis bullosa. Eur J Dermatol. 2016; 25 Suppl 1:30-2. https://doi.org/10.1684/ejd.2015.2552 PMid:26083672
8. Richards D. Malignant transformation rates in Oral Lichen Planus. Evid Based Dent. 2018; 19(4):122. https://doi.org/10.1038/sj.ejbd.6401349 PMid:30573869
9. Singh SK, Saikia UN, Ajit C, Kumar B. Squamous cell carcinoma arising from hypertrophic lichen planus. J Eur Acad Dermatol Venereol. 2006; 20(6):745-6. https://doi.org/10.1111/j.1468-3083.2006.01500.x PMid:16836514
10. Sigurgeirsson B, Lindelöf B. Lichen planus and malignancy. An epidemiologic study of 2071 patients and a review of the literature. Arch Dermatol. 1991; 127(11):1684-8. https://doi.org/10.1001/archderm.1991.01680100084009 PMid:1952973
11. Friedl TK, Flajg MJ, Ruzicka T, Rupec RA. Verruköses spinozelluläres Karzinom an Lichen ruber hypertrophicus. Drei Fallberichte mit Literaturüberblick. Hautarzt. 2011; 62(1):40-5. https://doi.org/10.1007/s00105-010-1961-4 PMid:20664992
12. Haenen CCP, Buurma AAJ, Genders RE, Quint KD. Squamous cell carcinoma arising in hypertrophic lichen planus. BMJ Case Rep. 2018; 2018. https://doi.org/10.1136/bmjcaserep-2017-002440 PMid:30002207
13. Desroche T, Moawad S, Cuny JF, Lerondeau B, Simon E, Burszttein AC, Schmutz JL. Bilateral squamous cell carcinoma arising from cutaneous ulcerative lichen planus of the extremities: two case reports. Eur J Dermatol. 2018; 28(3):391-392. https://doi.org/10.1684/ejd.2018.3255 PMid:29547127
14. Ali NM, Bhat R, Rao SB. Concurrent Presentation of Erythrodermic Lichen Planus and Squamous Cell Carcinoma: Coincidence or Malignant Transformation? Indian J Dermatol. 2015; 60(6):457-60. https://doi.org/10.4103/0019-1964.154355 PMid:26538691 PMCID:PMC4601411
15. Ghosh S, Kotele S, Ananda Rao PB, Turlapati SP, Kumar Soren D. Squamous cell carcinoma developing in a cutaneous lichen planus lesion: a rare case. Case Rep Dermatol Med. 2014. https://doi.org/10.1155/2014/205638 PMid:24839562 PMCID:PMC4006557
16. Garrido Colmenero C, Martín Castro A, Valenzuela Salas I, Martínez García E, Blasco Morente G, Tercero Sánchez J. Squamous cell carcinoma in lichen planopilaris. J Dermatol Case Rep. 2013; 7(3):84-7. https://doi.org/10.3315/jcder.2013.1147 PMid:24133362 PMCID:PMC3797015
17. Giesecke LM, Reid CM, James CL, Huiogl SC. Giant keratoacanthoma arising in hypertrophic lichen planus. Australas J Dermatol. 2003; 44(4):267-9. https://doi.org/10.1046/j.1440-0960.2003.00066.x PMid:14616493
18. Krasowska D, Bogaczewicz J, Chodorowska G. Development of squamous cell carcinoma within lesions of cutaneous lichen planus. Eur J Dermatol. 2007; 17(5):447-8.
19. Abdou AG, Marae AH, Shoibeh M, Dawood G, Abouelhath E. C-Jun expression in lichen planus, psoriasis, and cutaneous squamous cell carcinoma, an immunohistochemical study. J Immunooassay Immunochem. 2018; 39(1):58-69. https://doi.org/10.1080/15321819.2017.1395347 PMid:29144206
20. Bloor BK, Timdan N, Leigh IM, Odell E, Dogan B, Wolinia U, Ghalil M, Waseem A. Expression of keratin K2e in cutaneous and oral lesions: association with keratinocyte activation, proliferation, and keratinization. Am J Pathol. 2003; 162(3):963-75. https://doi.org/10.1016/S0002-9440(03)03891-8