CASE REPORT

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Oral Squamous Cell Carcinoma arising from Lichenoid Contact Reaction: A Case Report

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Abstract

Lichenoid contact reaction is a type of delayed hypersensitivity reaction to constituents derived from dental materials. It may occasionally transform into oral squamous cell carcinoma. A 62-year-male reported with the chief complaint of a wound in the inner aspect of right cheek for 2 weeks. On initial examination, a diffuse erythematous area with interspersed non-scrapable white striae was present on the right buccal mucosa and residual alveolar ridge with respect to 47 extending till gingivobuccal aspect of 45, roughly linear in shape, approximately 3x1 cm^2 in size, with a rough overlying surface. Ointment triamcinolone 0.1% was prescribed for 1 week. The patient visited after 3 months. Clinical examination revealed an ulcer with peripheral induration of size approximately 2.5 mm on right buccal mucosa adjacent to 46. An incisional biopsy revealed oral squamous cell carcinoma. Long-term follow-up of patients with oral lichenoid reaction is mandatory for early detection of malignant transformation.

Keywords: Lichen planus oral; Squamous cell carcinoma of head and neck; Ulcer.

Declarations

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Oral lichenoid reactions (OLRs) are chronic inflammatory lesions of the oral mucosa that occur as an allergic response to dental materials, certain medications and in patients with graft-vs-host disease (GVHD) [1]. Oral lichenoid contact reaction (OLCR) is considered to be due to a delayed (contact) hypersensitivity reaction to constituents derived from dental materials such as amalgam, gold, composites, and glass ionomers [2]. A higher proportion of women are affected by OLCR with a prevalence of 2.4% in the general population [3]. The majority of OLCRs are confined to sites that are regularly in contact with dental materials, such as the buccal mucosa and the border of the tongue, and present as white striations, plaques, erythema, ulcers, or blisters. The patients usually complain of sensitivity to spicy foods or burning sensation [4]. OLCR are those disorders that do not present the clinical and/or histopathological criteria considered typical with oral lichen planus (OLP) as mentioned in the Modified World Health Organization (WHO) diagnostic criteria for OLP [5]. The most apparent clinical difference between OLP and OLCR is the fact that OLCR are asymmetric and usually unilateral, making this a dominant differential distinction [6]. Histopathology will not normally be of much help in discrimination between OLP and OLCR but may be useful in identifying dysplasia and any risk of malignant transformation [4]. We present a case of malignant transformation of OLCR into oral squamous cell carcinoma (OSCC).

CASE

A 62-year-old male reported to the Department of Oral Medicine and Radiology of B. P. Koirala Institute of Health Sciences, Dharan with the chief complaint of a wound in the inner aspect of the right cheek region for 2 weeks. On examination, a diffuse erythematous area was present with interspersed non-scrapable white striae on right buccal mucosa and residual alveolar ridge with respect to 47 extending till gingivobuccal aspect of 45, roughly linear in shape, approximately 3 cm x 1 cm in size, with a rough overlying surface. The erythematous area was associated with a burning sensation with hot and spicy food, tender on palpation with no discharge and no peripheral induration **(Fig. 1)**. He had visited a general practitioner two years ago for the same problem and ointment triamcinolone 0.1% (Kenacort) was prescribed. It resulted in partial remission and later recurrence was seen.

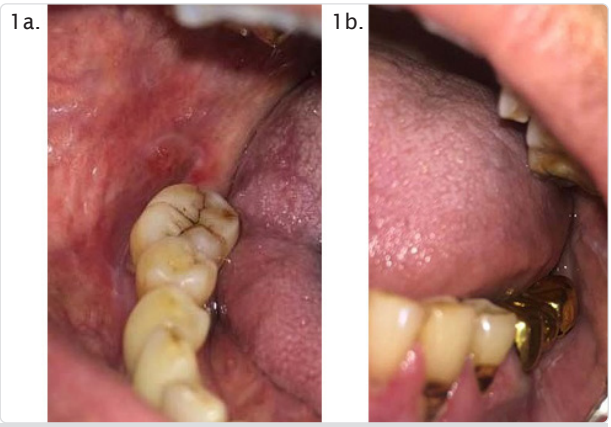

Figure 1: a. Initial presentation, diffuse erythematous area with interspersed non-scrapable white striae present on the residual alveolar ridge with respect to 47 extending till gingivobuccal aspect of 45. b. White radiating linear striae on erythematous background present on the left buccal mucosa extending along the gingivobuccal sulcus from distal aspect of 37 till 36.

Similarly, white radiating linear striae on the erythematous background were present on the left buccal mucosa extending from the distal aspect of 37 till 36 along the gingivobuccal sulcus, size approximately 2.5 cm x 1 cm, roughly linear in shape. The surrounding mucosa appeared normal. It was non-tender on palpation without any discharge and peripheral induration was not present. There were no similar lesions elsewhere in the body. There was metal crown (gold) restoration on 14, 15 and 16 and 34, 35 and 36 and none of the teeth were carious. There was a close contact of metal crown restoration with buccal vestibule adjacent to 45, 46, 47 regions on centric occlusion and centric relation. On extra-oral examination, there was no cervical lymphadenopathy.

The patient was a known hypertensive, on medication and did not have a history of smoking or alcohol consumption but had a habit of chewing betel nut (pan) since last 2 years, which he had quitted 6 months ago. The complete blood count, thyroid function test, and random blood sugar were done, and reports were within normal limits. Serology for HIV, HCV, HBsAg were non-reactive. Based on the clinical findings, the tentative diagnosis was bilateral OLCR and differential diagnoses of OLP, oral lichenoid drug eruption, discoid lupus erythematous eruptions were made. An ointment triamcinolone 0.1% (Oroheal) was prescribed for 1 week.

The patient was advised for follow-up in 1 week and planned for the metal crown removal. However, the patient visited after 3 months. Three months follow-up

visit revealed ulcer with peripheral induration of size approximately 2.5 mm on right buccal mucosa adjacent to 46 **(Fig.2)**. The intra-oral periapical radiograph with respect to 46 revealed horizontal bone loss with respect to 45 and 46 of approximately 2 mm **(Fig. 3)**.

Incisional biopsy was planned and performed from right buccal mucosa adjacent to 46 in aseptic condition under local anesthesia and sent for the histopathological examination **(Fig. 4)**. The histopathologic examination revealed islands and strands of epithelial tumor cells in the underlying connective tissue showing features of epithelial dysplasia in the form of cellular and nuclear pleomorphism, nuclear hyperchromatism and dyskeratosis. The connective tissue was fibrocellular with inflammatory cells infiltrates predominantly lymphocytes and plasma cells along with numerous endothelial cells lined with blood vessels of variable size. The histopathological findings were suggestive of the oral squamous cell carcinoma **(Fig. 5)**. The patient was referred to an oncological center for further treatment where contrast enhanced computed tomography was done.

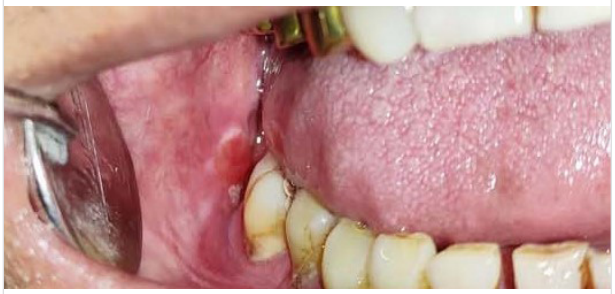

Figure 2: A single, well-defined ulcer with peripheral induration with erythematous area with respect to 46.

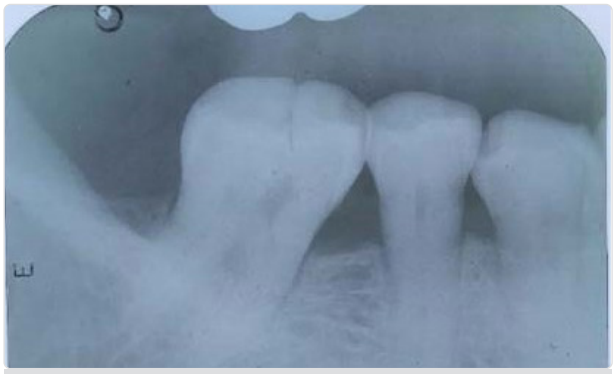

Figure 3: Intraoral periapical radiograph with respect to 46.

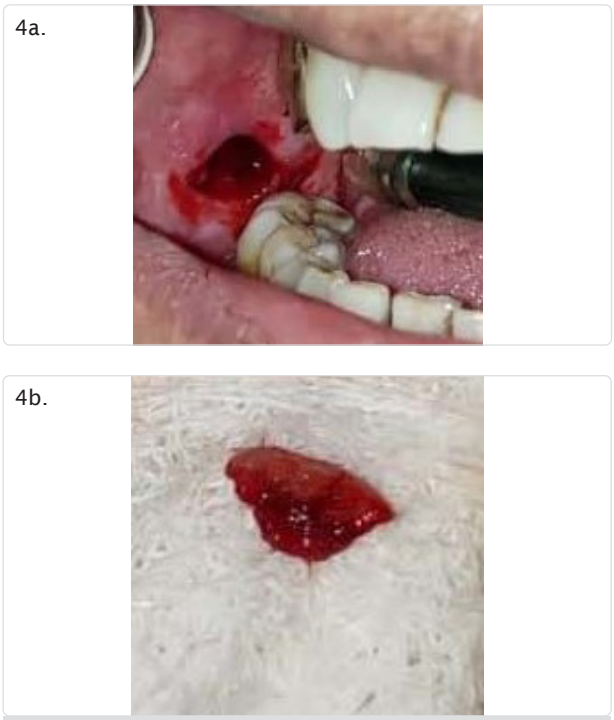

Figure 4: Biopsy specimen.

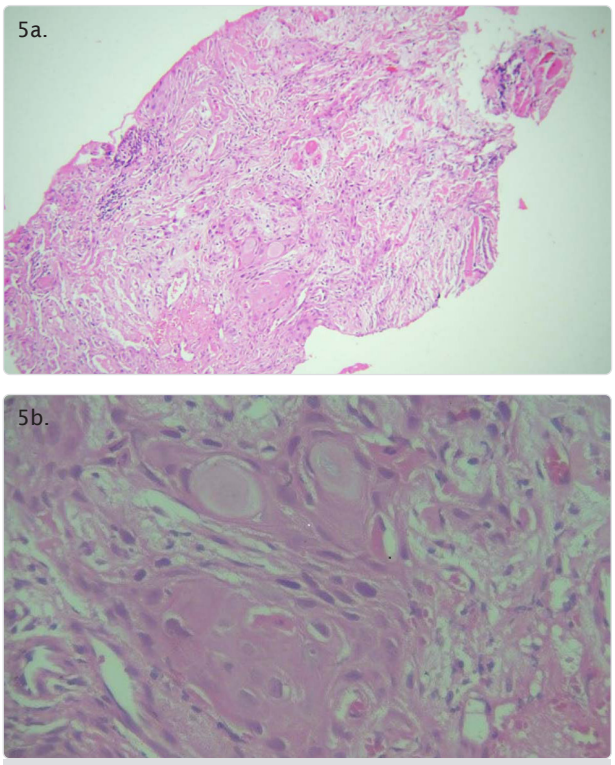

Figure 5: Photomicrographs of oral squamous cell carcinoma; H and E: a. 10x revealed islands and strands of epithelial tumor cells in the underlying connective tissue showing the features of epithelial dysplasia. b. 40x magnification revealed cellular and nuclear pleomorphism, nuclear hyperchromatism and dyskeratosis.

DISCUSSION

The WHO Collaborating Centre for Oral Cancer has classified oral lichenoid reaction as oral potentially malignant disorders (OPMD) as there was sufficient evidence for an increased risk of developing into squamous cell carcinoma among patients diagnosed with OLR [7]. OLR are histologically indistinguishable from OLP but unlike OLP could be etiologically related to causative agents such as medications, systemic diseases, or dental materials [8]. OLCR is most often attributable to dental restorative materials, most commonly amalgam. With the removal and replacement of the putative causative material, the majority of OLCRs resolve within one to two months [3].

The characteristic feature of OLCR is the lesion's direct topographic relationship to the suspected causative agent and typical sites include the lateral borders of the tongue and the buccal mucosa having a direct anatomical relationship with the filling/dental restoration(s). As OLP and OLCR are indistinguishable clinically and histologically, they are grouped under the term oral lichenoid lesions (OLL). The high frequency of contact allergy to gold in patients with OLL was found to suggest that dental gold can be one of several etiological factors for developing or maintaining OLL. Thus, restoration with dental gold should be avoided in OLL patients [9].

In OLCR, lesions are limited to such contacts, whereas in lichen planus the lesions may affect oral mucosal sites not in contact with restorations, the gingivae, or other mucocutaneous sites, e.g. skin or vulvo-vaginal mucosa [10]. The malignant transformation rate is 0.5% to 2% in OLP whereas 2% to 4% in LCRs [11].

A higher proportion of women are affected by OLCR [3]. Treatment consists of removal/ replacement/ coverage of restorations that are in direct physical contact with mucosal lesions. OSCC development could reflect either a stochastic event driven by chance or, alternatively, it may be speculated that genetic aberrations occurring before clinical intervention could persist and progress to cancerization even in apparently normal mucosa [8]. The cause of increased oral cancer risk in OLP patients is not because OLP is inherently pre-malignant but because the oral mucosa affected by OLP may be more sensitive to 'exogenous mutagens' [12].

The same reasoning applies well to LCR lesions as the histopathological nature of these reactions are the same as OLP. The gradual development of dysplasia or candida infection may have interfered with the original LCR-type lesions in the period preceding the development of OSCC. Some of the LCR lesions may have appeared initially as part of 'idiopathic' OLP. The inherent properties paved the way for further and final malignant changes due to exogenous dental material-associated factors impinging upon the lesion. Thus, the association between material-associated LCR and the development of OSCC should not be clinically ignored [13].

Widespread red lesions in the oral cavity are usually left undiagnosed and untreated unless they turn symptomatic. It is due to the lack of public awareness and knowledge. Any red lesion in the oral cavity should be evaluated with a proper history from the patient at the earliest and planned for a biopsy immediately [14].

CONCLUSION

Considering the risk of malignant transformation with oral lichenoid reaction, even if asymptomatic or barely symptomatic, scrupulous long-term follow-up and histopathological examination at more frequent intervals from the time of the first diagnosis is mandatory.

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