Malignant transformation of oral lichen planus in lingual location: report of a case

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Summary  One of the most controversial aspects of oral lichen planus (OLP) is its malignant potential. There have been a few well-documented reports of malignant transformation in the absence of known exposure to exogenous carcinogens. The aim of this study is to report the case of a patient with a lesion previously diagnosed as oral lichen planus, who developed squamous cell carcinoma in the same location, in the absence of known exposure to exogenous carcinogens.

KEYWORDS  Oral lichen planus; Squamous cell carcinoma; Malignant transformation

Introduction

Lichen planus is a chronic mucocutaneous inflammatory disease of unknown etiology, frequently occurring in oral locations, with distinctive though not entirely diagnostic clinical and histopathological characteristics.

One of the most controversial aspects of oral lichen planus (OLP) is its malignant potential. Various studies have suggested that OLP has potential for malignant transformation, although there is consensus that the probability of such transformation is low. There have been a few well-documented reports of malignant transformation in the absence of known exposure to exogenous carcinogens.

Here, we report the case of a patient with a lesion previously diagnosed as oral lichen planus, who developed squamous cell carcinoma in the same location, in the absence of known exposure to exogenous carcinogens.

Case report

A 45-year-old woman who in June 1995 was referred to the Oral Medicine Unit of the Dentistry Faculty of the University of Santiago de Compostela for evaluation of a red and white lesion in lingual location. Oral examination revealed a reddish atrophic lesion with a small central erosion.
surrounded by raised whitish streaks, approximately 2 cm in diameter, located on the dorsal surface of the right half of the tongue (Fig. 1). In addition, atrophic zones with whitish streaks were present on the alveolar mucosa of the upper jaw. The patient reported itchiness in tongue and gums, first noted about 3 months previously. She was a non-smoker and moderate social drinker with no relevant antecedents. Following a clinical diagnosis of atrophic-erosive lichen planus, biopsies were taken for histopathological studies. These revealed (a) stratified squamous epithelium with areas of acanthosis and hyperkeratosis, (b) the presence of a dense lymphocytic inflammatory infiltrate along the epithelium/connective-tissue interface, (c) hydropic degeneration of the basal layer, and (d) some necrotic keratinocytes (Fig. 2). Dysplasia was not observed. Immunofluorescence tests for detection of IgG, IgA, IgM, IgE and C3 were negative. The diagnosis of atrophic-erosive lichen planus was thus confirmed, and treatment was commenced with 0.3% aqueous triamcinolone acetonide as mouthwash, three times daily for 15 days. At next appointment the patient showed symptom improvement, but examination revealed erosive zones, so that we commenced topical treatment with 0.05% clobetasol propionate in ointment, three times daily for another 15 days. This examination also revealed that the ulcerated areas had disappeared, leaving an atrophic epithelium with whitish streaks. After 15 days, the treatment was stopped. The patient returned for regular check-ups every 3 months, showing alternating lesion recurrence and remission on treatment. In June 1998 the patient stopped attending check-ups.

In October 2002, the patient consulted again with a whitish exophytic lesion on the back of the tongue. Histopathological examination revealed (a) stratified flat epithelium showing acanthosis and slight hyperkeratosis. Note also the hydropic degeneration of the basal layer, and the dense lymphocytic inflammatory infiltrate along the epithelium/connective-tissue interface. Dysplasia is not seen. (Hematoxylin-eosin, 10×). (b) Higher-magnification of the micrograph in (a), showing the hydropic degeneration of the basal layer, and the inflammatory infiltrate along the epithelium/connective-tissue interface. Some necrotic keratinocytes can be seen. (Hematoxylin-eosin, 40×).

Figure 1 (a) Dorsal surface of the patient’s tongue, showing atrophic-erosive zones surrounded by whitish streaks. (b) Detail of Fig. 1a, showing an atrophic-erosive lesion on the dorsal surface of the right half of the tongue. Note the whitish streaks with reticular distribution, characteristic of lichen planus.

Figure 2 (a) Stratified flat epithelium showing acanthosis and slight hyperkeratosis. Note also the hydropic degeneration of the basal layer, and the dense lymphocytic inflammatory infiltrate along the epithelium/connective-tissue interface. Dysplasia is not seen. (Hematoxylin-eosin, 10×). (b) Higher-magnification of the micrograph in (a), showing the hydropic degeneration of the basal layer, and the inflammatory infiltrate along the epithelium/connective-tissue interface. Some necrotic keratinocytes can be seen. (Hematoxylin-eosin, 40×).
tongue, with areas of redness and ulceration, approximately 3 cm in diameter (Fig. 3). Biopsy revealed squamous cell carcinoma (Fig. 4), at clinical stage I (T2 N0 M0). The patient was referred to the Maxillofacial Surgery Service of the University Clinical Hospital of Santiago de Compostela, where the lesion was removed and the patient underwent radiotherapy (total dose 70 Gy, in a megavoltage cobalt-60 unit). The patient currently shows no sign of relapse, although radiotherapy has led to an oral mucositis of difficult management.

Discussion

The World Health Organization (WHO) defines oral lichen planus as a precancerous condition, associated with an increase in the risk of oral cancer. As noted, various authors have suggested that OLP has malignant potential, but other authors have disputed this view. In this connection, Krutchkoff et al. in a review of 223 cases published between 1950 and 1976, pointed out that only 15 met strict requirements for consideration as cases of malignant transformation of an existing OLP lesion.

More recently, van der Meij et al. have published a meta-analytical review of articles dealing with malignant transformation of oral lichen planus in the English-language literature over the period 1977–1998. Of the 98 documented cases, the authors found that only 33 (34%) met the criteria of Krutchkoff et al. Of the 65 cases that did not meet these criteria, 20 were inadequately documented as regards histopathology, one as regards clinical characteristics and course, 33 as regards both histopathology and clinical characteristics; in addition, four had been followed up for less than 2 years, and seven patients were smokers.

Here we have reported a case in which the patient developed a squamous cell carcinoma in the same location as a lesion diagnosed 7 years previously as OLP. Clinical appearance was an isolated single unilateral lesion, with absence of bilateral lesions in the jugal mucosa. This is not the typical presentation of OLP; however, the diagnosis was confirmed by the combination of the clinical appearance (atrophic lesion surrounded by raised whitish streaks) and histopathological findings in line with those characteristic of OLP.

Some researchers have suggested that these atrophic-erosive forms predispose the oral mucosa to the effects of other carcinogenic agents; as far as we know, however, our patient does not have a history of exposure to any major exogenous carcinogen. In any case, malignant transformation does not appear to be exclusive to atrophic-erosive
OLP: Silverman et al.,\textsuperscript{15} in a study of 214 cases of OLP followed up on average for 7.5 years, found five of malignant transformation, three affecting erosive lesions, one affecting an atrophic lesion, and one affecting a reticular lesion. Lo Muzio et al.\textsuperscript{16} reported 14 cases of squamous cell carcinoma associated with OLP lesions, which in 12 of the cases were plaque-like.

The body-location statistics of OLP-associated squamous cell carcinomas (SCCs) are of course different from those of SCCs in general. SCCs on the back of the tongue are very infrequent, different from those of SCCs in general. SCCs on the tongue.

However, malignant transformation of OLP in this location is relatively frequent, and some authors have suggested that this location is a significant risk factor.\textsuperscript{18}

Malignant transformation of OLP appears to be independent of exogenous risk factors. Thus Garcia-Pola et al.\textsuperscript{19} reported 4 cases of SCC in the same location as OLP lesions, but only in one of these cases was the patient a smoker. Similarly, in the present case (in which the OLP was conclusively diagnosed, and in which we confirmed that the SCC developed in exactly the same location as the OLP) the patient had no history of exposure to exogenous carcinogens. The present case thus supports the view that OLP may undergo malignant transformation, and that this does not require exogenous carcinogens. In conclusion, despite the current uncertainty about the probability of malignant transformation of OLP, the fact that such transformation may occur seems increasingly certain.\textsuperscript{16,19,20} This argues for a need to perform routine monitoring of some subsets of OLP patient, including patients with atrophic-erosive lesions on the tongue.

References