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Case history

Malignant Transformation of Labial Lichen Planus: A Case Report and a Review of the Literature

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Abstract

Oral Lichen Planus (OLP) is a chronic inflammatory dermatosis usually benign that can affect skin, integuments and mucosal membranes presented with various clinical appearances. Typically characterized by the presence of white lace-like lesions, with or without atrophic or erosive areas. Its diagnosis is based on the clinical examination and histological analysis. In most of the cases, the OLP has a benign evolution but malignant transformation may occur justifying the strict surveillance of the disease and effective treatment of relapses.

We report the case of a 58-year-old man, without specific and non-smoking history, reported to the Oral Surgery Department of the Consultation Center of Dental Treatment of Rabat, presenting oral lesions lasting for three years. The intraoral examination and biopsy were concluded an erosive oral lichen planus. Two years after, the labial lesion changed its appearance. Pathological diagnosis confirmed the presence of squamous cell carcinoma. Total surgical excision was performed with respect of carcinologic resection margins. The present case supports the view that OLP may undergo malignant transformation, and that this does not require exogenous carcinogens.

Oral lichen planus is a potentially malignant disorder with a capacity, although low, for malignant transformation. Of all the factors related to the process of malignant transformation, the chronic inflammation seems to be the key factor in the development of oral cancer. The highest rate of malignancy was noted in erythematous and erosive lesions. In this way, follow-up of OLP patients could be carried out more efficiently and appropriately.

Keywords: ***Oral lichen planus, malignant transformation, squamous cell carcinoma, oral cancer.***

Introduction

Oral lichen planus (OLP) is an autoimmune disease with an inflammatory origin and chronic progression [1] that affects 1–2% of the general adult population, mostly woman, usually aged between 30 and 60 years [2]. OLP can affect several sites of oral cavity, especially buccal mucosa, back of the tongue and its margins, gingival and labial mucosa [3]. That can manifest in a number of ways. The most common is reticular type which has a white lacy appearance, other forms include erosive, atrophic and plaque forms. For many patients, OLP considerably limits their essential daily activities such as eating, drinking, talking or interacting with others. Today OLP is classified as an oral potentially malignant disorder. The rate of OLP malignant transformation in Oral squamous cell carcinoma (OSCC) remains debated; however, the vast majority of studies reported a short follow-up [4,5], then this could induce to underestimate the problem. 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. We report the case of a 58-year-old man, without specific and non-smoking history, reported to the Oral Surgery Department of the Consultation Center of Dental Treatment of Rabat, presenting oral lesions lasting for three years.

Case History

A 58 years-old man was referred in 2014 to the Oral Surgery Department of the Consultation Center of Dental Treatment of Rabat for evaluation of buccal, labial, lingual and gingival erosive lesions evolving for three years. He had consulted a dermatologist, who had prescribed topical antifungal and mouthwash, which had resulted in no significant changes in disease process. He complains of a burning pain in the oral cavity on eating and drinking. Without any specific or smoking history. Oral examination revealed bilateral erosive lesions extending over labial, lingual and buccal mucosa (**Figure 1 and 2**). Erosive labial lesion with keratotic plaque (**Figure 3**) and generalized gingival inflammation were noted. Lesions were clinically diagnosed as erosive oral lichen planus. A biopsy was taken from lower labial mucosa. The diagnosis of erosive lichen planus was thus confirmed and treatment was instaured with topical steroids as mouthwash three

times daily (solupred®).This patient had discontinued his regular follow-up outside in our service. In March 2016, the patient presented to our service complaining of pain and recurrence of lesions after cessation of treatment. Lesions have changed in appearance in the lower lip with the occurrence of whitish elevations; lesions stand out and showed an erosive appearance. A biopsy was carried out and concluded a micro-infiltrating squamous cell carcinoma (**Figure 4**). The patient was referred to the maxillofacial surgery department for an adequate surgical management of the malignant lesion. He benefited from a carcinological excision taking the entire red lip, whose reconstruction of which was made by two mucous flaps (**Figure 5**). Patient was invited to strict follow-up to detect local recurrence or second primary carcinoma.

Figure 1: (a and b): Diffuse erosive lesion of the labial and buccal mucosa associated with keratotic areas

Figure 2: Atrophic lichen planus of the dorsal and lateral tongue (anterior ulceration is the seat of a biopsy)

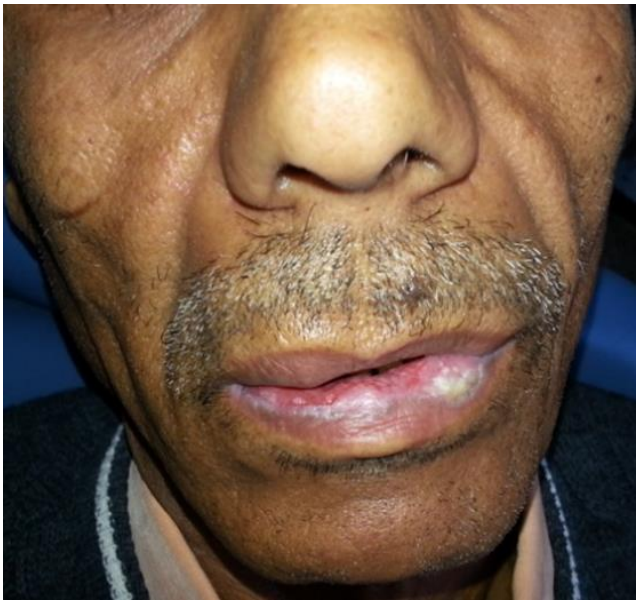

Figure 3: White keratotic plaque and erosive lesion on the vermillion

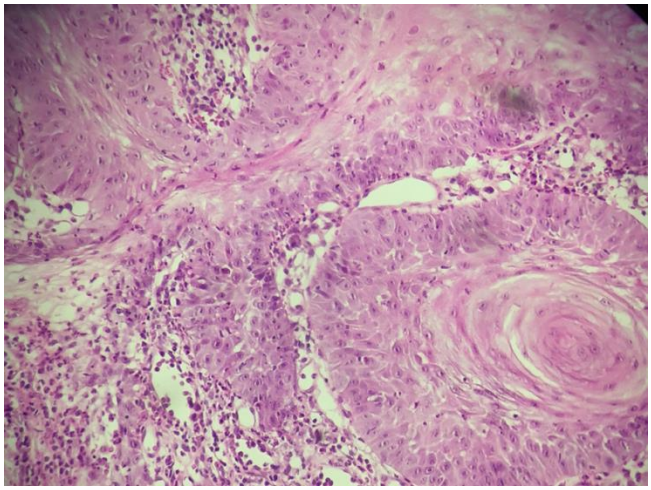

Figure 4: histological aspect in favor of a micro-invasive Squamous Cell Carcinoma

Figure 5: The patient after lip excision and reconstruction by two mucosa flaps

Discussion

In 2005 the World Health Organization (WHO) classifies OLP as a "potentially malignant disorder" with unspecified malignant transformation risk [6]. His true incidence of OLP malignant transformation remains controversial and results are somewhat difficult to interpret because of the heterogeneity of the study populations [7]. The reported frequency of malignant transformation ranges from 0% to 12.5% [8]. Giuliani et al. conducted a systematic review to determine the malignant transformation rate of OLP and oral lichenoid lesion (OLL) and their risk factors [9]. He found an overall transformation rate of 1.40% (1.37% for OLP and 2.43% for OLL) and an annual transformation rate of 0.20%. Which was comparable with other recent meta-analyses which obtained combined values of 1.09% [10] and 1.1%, respectively [11]. Interestingly, when a sub-group analysis was performed utilizing only studies adhering to the updated 2003 WHO criteria, which specifically excludes lesions with dysplasia, Aghbari et al [11] obtained a malignant transformation rate of 0.9%. This highlights the importance of lesion classification, as grade of dysplasia has been correlated with malignant transformation malignant transformation risk, and studies that consider that OLP can include dysplasia may have artificially high rates [12]. In order to properly investigate the potential for malignant transformation of OLP long term multicentre cohort studies with good clinical and histological information are needed [9].

A recent meta-analysis reported that 1.1% of OLP lesions progress into OSCC with a higher incidence in smokers, alcohol users, and in those infected with hepatitis C virus (HCV) [11]. However, our patient does not have a history of exposure to any major exogenous carcinogen or HCV infection. The present case thus supports the view that OLP may undergo malignant transformation, and that this does not require exogenous carcinogens. Thus Garcia-Pola et al. reported 4 cases of SCC in the same location as OLP lesions, but only in one of these cases was the patient a smoker [13,14]. Multiple studies have shown that female gender, erosive forms and tongue site should be considered as risk factors for OLP transformation [9]. While other authors consider the atrophic-erosive lesions as having the highest rate of malignant transformation [6]. Some studies also mention a relevancy of keratotic forms (plaques), either when appearing alone, or when associated with atrophic erosive lesions [6]. Mattsson et al [15] reported that the transformation of this disease cannot be explained by specific clinical features, since different types of OLP showed a similar percentage of transformation. The pathogenesis of OLP and the mechanism for malignant transformation are still unclear. The prevailing theory proposes that chronic stimulation by inflammatory cells lead to an alteration growth of keratinocytes, and with oxidative stress, deoxyribonucleic acid (DNA) damage results in neoplastic change [16].

OLP alternates between periods of remission and exacerbation, thus a scheduled follow-up is strongly recommended [10]. Treatment is normally reserved only for symptomatic patients. Topical steroid therapy is generally administered, although most physicians accept the hypothesis that the maintenance of this therapy may inhibit the chronic inflammatory processes present in OLP, particularly the severe inflammation noted in erosive disease, and in this way prevent the progression to malignant transformation [17].

List of abbreviations

OLP: Oral Lichen Planus
OSCC: Oral Squamous Cell Carcinoma
WHO: World Health Organization
OLL: Oral Lichenoid Lesion
HCV: hepatitis C virus
SCC: Squamous Cell Carcinoma
DNA: deoxyribonucleic acid

Conflicts of Interest

There is no conflict of interest regarding the publication of this paper.

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Authors' contributions

HF clinical case follow-up and manuscript writing. CS clinical case follow-up and manuscript writing. ZY English translation of the manuscript. All authors read and approved the final manuscript.

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