Objective: The objective of this study was to report a case of oral leukoerythroplakia, which is a potentially malignant disorder and has a high malignant transformation rate.

Method: A 58 year old male patient reported with the chief complaint of burning sensation on his right inner cheek region. On clinical examination he was diagnosed as a case of oral leukoerythroplakia and excisional biopsy was performed.

Results: Excisional biopsy revealed a highly dysplastic atrophic parakeratinized epithelium with dense inflammatory infiltrate, confirming the clinical diagnosis of oral leukoerythroplakia.

Conclusion: All mixed red lesions should be examined carefully since many of these could turn out to be oral leukoerythroplakia.

Keywords: Erythroplakia, Leukoerythroplakia, Speckled, Pre-malignant.

INTRODUCTION

The term oral erythroplakia is used to describe a red plaque or macular lesion in the mouth for which a specific clinical diagnosis cannot be established. Lesions are named erythroleukoplakia, leukoerythroplakia or speckled leukoplakia when white patches are present over the red plaque.1 Erythroplakia is a clinical term; it does not indicate a particular microscopic diagnosis, although after a biopsy most are found to be severe dysplasia or carcinoma.2 In very high risk cases, such as oral floor lesions in heavy smokers and alcohol abusers, 80% of these red patches may already contain focal areas of microinvasive cancer at the time of initial biopsy. Its usual microscopic counterpart, carcinoma in situ, has been shown to recur and transform into invasive carcinoma in approximately 25% of treated cases.3 The objective of this study was to report a case of oral leukoerythroplakia, which is a potentially malignant disorder and has a high malignant transformation rate.
CASE REPORT

A 58 year old male patient reported to our department with the chief complaint of burning sensation on his right inner cheek region. The patient had been aware of a red patch on his right inner cheek for about 2 years; however, he had not sought any treatment as the lesion was not painful. Medical and dental histories were non-contributory. Patient had the habit of smoking of bidi approx 25bidis/day and occasional alcohol consumption. Intraoral examination showed 2cm X 1.5cm bright red patch with white specks on its surface on the right buccal mucosa (Fig 1). The lesion was sharply demarcated from the surrounding normal mucosa. No surrounding induration was present. Clinical diagnosis of leukoerythroplakia was made. Complete surgical excision of the lesion was advised (Fig 2). Histopathological examination of the biopsy showed highly dysplastic, atrophic parakeratinized stratified squamous epithelium with dysplastic features like nuclear hyperchromatism, increased nuclear cytoplasmic ratio, nuclear and cellular pleomorphism and few mitotic figures. Underlying connective tissue stroma showed dense inflammatory cell infiltrate, chiefly lymphocyte. Based on histopathological features, a diagnosis of severe dysplasia was made (Fig 3).

DISCUSSION

In 1911, Queyrat described a sharply defined, bright red, glistening velvety lesion of the glans penis, which was termed ‘erythroplasie’. Although red lesions of the oral mucosa have been noted for many years, the use of the term “erythroplakia” in this context has been common for only about 25 years.4

Over the years several definitions for oral erythroplakia have been suggested. Most accepted is the one given by WHO, which describes it as, “any lesion of the oral mucosa that presents as bright red velvety plaques which cannot be characterized clinically or pathologically as any other recognizable condition”.5 An updated definition for erythroplakia was proposed by Bouquot as “a chronic red mucosal macule which cannot be given another specific diagnostic name and cannot be attributed to traumatic, vascular, or inflammatory causes”. Erythroplakia patches may be located near, or associated with, oral leukoplakias. Bouquot and Whitaker suggested that erythroplakia may occur with leukoplakia in the stage called erythroleukoplakia.6

The prevalence rate of these lesions has been reported between 0.01%- 0.21%. The incidence is not known, but the average annual incidence for microscopically proven oral carcinoma in situ, which represents the great majority of erythroplakias, has been estimated to be 1.2 per 100,000 population (2.0 in males and 0.5 in females) in the United States.7

It predominately occurs in the floor of the mouth, buccal mucosa, soft palate, ventral tongue and tonsillar fauces.8 In a study on 58 cases of erythroplakia, the disease was found to be more common among people in their 50s and 60s. The risk factors for oral cancer such as chewing tobacco, smoking, and alcohol drinking are assumed to be associated with erythroplakia. In a recent case-series study, erythroplakia was associated with a high prevalence of TP53 mutations. TP53 mutations may be associated with tobacco exposure for oral cancer, which would possibly indicate that tobacco exposure may play an important role in the development of erythroplakia.6

The differential diagnosis includes: erythematous candidiasis, early SCC, local irritation, mucositis, lichen planus, lupus erythematosus, drug reaction and median rhomboid glossitis.8
 localized areas of redness are not uncommon in the oral cavity, areas of erythroplakia are likely to be disregarded by the examiner, and they are often falsely determined to be a transient inflammatory response to local irritation. Differentiation of erythroplakia from benign inflammatory lesions of the oral mucosa can be enhanced by the use of a 1% solution of toluidine blue, applied topically with a swab or as an oral rinse.

Histopathologically, epithelium shows lack of keratin production and is often atrophic, but it may be hyperplastic. This lack of keratinization and epithelial thinness allows the underlying microvasculature to show through, thereby causing the red color. Epithelium shows dysplastic features like hyperchromatism, pleomorphism and increase in number of mitotic figures. In a sister study, to their large series of leukoplakia cases, Shafer and Waldron also analyzed their biopsy experience with 65 cases of erythroplakia. All the erythroplakia cases showed some degree of epithelial dysplasia; 51% showed invasive squamous cell carcinoma, 40% were carcinoma in situ or severe epithelial dysplasia, and the remaining 9% demonstrated mild-to-moderate dysplasia. Therefore, true clinical erythroplakia is a much more worrisome lesion than leukoplakia.

Erythroplakia has been considered the most severe form among all of the oral premalignant lesions because of its high malignant potential. Generally, transformation rates, including those with invasive carcinoma already at biopsy, vary from 14% to 50%. Table 1 shows the malignant transformation of various premalignant lesions.

The treatment of choice for erythroplakia is surgical excision. It is generally more important to excise widely than to excise deeply in dysplastic and in situ lesions because of their superficial nature and the fact that dysplastic cells usually extend beyond the clinically evident lesion. However, since recurrence and multifocal involvement is common, long-term follow-up is mandatory.

CONCLUSION

Erythroplakia has been called "the dangerous oral mucosa" because it typically presents as carcinoma in situ, severe epithelial dysplasia or superficially invasive carcinoma under the microscope. There is currently no unique reliable parameter to identify these lesions predictive of malignant transformation. Risk assessment is usually based on clinical, pathological and more recently on bio-molecular evaluations. Few data are available on oral erythroplakia and there is an urgent need for randomized controlled trials.

REFERENCES


Table 1: Potentially malignant lesions of the oral, pharyngeal and laryngeal mucosa, clinical terms only. 3

<table>
<thead>
<tr>
<th>Disease name</th>
<th>Malignant transformation potential</th>
</tr>
</thead>
<tbody>
<tr>
<td>Proliferative verrucous leukoplakia</td>
<td>*****</td>
</tr>
<tr>
<td>Erythroplakia</td>
<td>*****</td>
</tr>
<tr>
<td>Nicotine palatinus in reverse smokers</td>
<td>*****</td>
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<tr>
<td>Oral submucous fibrosis</td>
<td>****</td>
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<tr>
<td>Speckled, granular (non-homogeneous) leukoplakia</td>
<td>****</td>
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<tr>
<td>Laryngeal keratosis/ leukoplakia</td>
<td>***</td>
</tr>
<tr>
<td>Actinic cheilitis</td>
<td>***</td>
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<tr>
<td>Smooth, thick (homogeneous) leukoplakia</td>
<td>**</td>
</tr>
<tr>
<td>Smokeless tobacco keratosis</td>
<td>*</td>
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<tr>
<td>Lichen planus</td>
<td>*</td>
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</tbody>
</table>

(*) indicates the severity of malignant transformation.
Figure No. 1: Photograph showing a red patch with white nodules on the buccal mucosa.

Figure No. 2: Complete excised lesion.
Figure No. 3: Photomicrograph showing atrophic parakeratized stratified squamous epithelium with severe dysplastic features. Dense inflammatory cell infiltrate is also seen.