Oral erythroleukoplakia – a potentially malignant disorder

Erytroleukoplakia jamy ustnej – schorzenie o charakterze potencjalnie złośliwym

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ABSTRACT

Erythroleukoplakia or speckled leukoplakia is a painless lesion characterized by an association of white areas interspersed with red areas, resistant to scratching and with potential for malignancy. It appears most frequently in the oral mucosa, vermilion and gums. The etiology of erythroleukoplakia is variable, but may be related to smoking, alcoholism, microorganisms, and other agents. The objective of this work was to report a case of erythroleukoplakia in a male patient who was a smoker and an alcoholic. The lesion was detected in the retrocomissural region during a routine dental examination. The diagnosis was established after histopathological examination of the tissue specimen collected by the excisional biopsy. Otolaryngologists and dentists performing an oral examination should be alert to the patient’s habit of smoking and drinking. Oral biopsy is mandatory to recognize the presence and the severity of epithelial dysplasia, which is a decisive factor for subsequent treatment planning.

KEYWORDS: Erythroleukoplakia, Leukoplakia, Smoking, Oral Mucosa.

STRESZCZENIE

Erythroleukoplakia or speckled leukoplakia is a painless lesion characterized by an association of white areas interspersed with red areas, resistant to scratching and with potential for malignancy. It appears most frequently in the oral mucosa, vermilion and gums. The etiology of erythroleukoplakia is variable, but may be related to smoking, alcoholism, microorganisms, and other agents. The objective of this work was to report a case of erythroleukoplakia in a male patient who was a smoker and an alcoholic. The lesion was detected in the retrocomissural region during a routine dental examination. The diagnosis was established after histopathological examination of the tissue specimen collected by the excisional biopsy. Otolaryngologists and dentists performing an oral examination should be alert to the patient’s habit of smoking and drinking. Oral biopsy is mandatory to recognize the presence and the severity of epithelial dysplasia, which is a decisive factor for subsequent treatment planning.

SŁOWA KLUCZOWE: Erythroleukoplakia, leukoplakia, nikotynizm, błona śluzowa jamy ustnej.

INTRODUCTION

Leukoplakia (leukos meaning white; plakia meaning patch) is a disease characterized by the presence of a white plate which constitutes an expression of alteration in the keratinization process [1]. In 1877, Schwimmer was the first author that used this term to describe a white plaque on the tongue [2]. However, it can also develop in the oral mucosa, penis or vulva. According to the World Health Organization, leukoplakia is described as a white lesion that cannot be removed by scraping. It is caused by local irritating factors (smoking and alcohol) which stimulate the formation of hyperkeratosis. The incisional biopsy is essential for diagnosis [1].

The natural history of oral leukoplakia remains poorly understood [4]. Oral leukoplakia can develop into a carcinoma. Malignant
transformation rates of oral leukoplakia range from 0.13 to 17.5%. The risk factors of malignant transformation in the buccal mucosa and labial commissure are male gender with chewing tobacco or smoking in some countries such as India, or older age and/or being a non-smoking female in other countries [5].

According to the clinical appearance, leukoplakia can be classified into four types: early or thin, homogenous or thick, granular or verruciform and speckled or erythroleukoplakia. Each subdivision has a different malignant transformation potential. For example, thin leukoplakia often becomes malignant without clinical changes. Thick leukoplakia undergoes malignant transformation in 1–7% of cases [6].

Histologically, leukoplakia can show several epithelial changes. The presence of dysplasia with varying degrees of severity is very common. A study developed by Waldron and Shafer demonstrated that 19.9% of leukoplakia cases had some degrees of dysplasia, 3.1% were frank carcinoma, 4.6% showed severe dysplasia or carcinoma in situ, and 12.2% showed mild to moderate dysplasia [7].

Erythroplakia is another oral lesion considered as a potentially malignant disorder which precedes the development of cancer. Erythroplakia is a red lesion characterized by patches, plaques or erosions of various dimensions that affect regions of the hard and soft palate, floor of the mouth, tongue and retromolar region. It can also be caused by local irritating factors, such as tobacco and alcohol [8]. Erythroplakia, when associated with leukoplakia, has a higher malignant potential and shall be called erythroleukoplakia [3]. In order to establish the diagnosis, it is also necessary to perform a biopsy and this should be done in the perilesional region. The histopathological features of erythroleukoplakia include epithelial atrophy with varying degrees of atypia [9].

The treatment of choice for oral leukoplakia and erythroplakia is surgery. There exists a possibility of recurrence so monitoring of the patient should be permanent. The first step in the treatment of oral leukoplakia and erythroplasias is advising the patient to stop his/her habits (alcohol and tobacco). The treatment of these lesions may be conservative or surgical [10]. Oral precancerous lesions can be eradicated by surgical excision [11], laser surgery [12], or photodynamic therapy [13]. The aim of this paper was to report on a case of erythroleukoplakia that developed bilaterally in the buccal mucosa of an alcoholic and smoker.

**CASE REPORT**

Male, 37 years old, presented to the clinic of the School of Dentistry (Universidade Federal do Paraná, Curitiba, Brazil) complaining of pain in the posterior teeth and difficulty in chewing.

Figure 1. Erythroleukoplakia - White plaque with red areas in the buccal mucosa

During the anamnesis, the patient reported tenderness in the posterior teeth and a temporo-mandibular joint pain. Physical examination revealed the presence of a removable partial denture that was not adapted. Dental caries, residual roots, periodontal disease, bad breath, poor oral hygiene, nicotine stomatitis, and smoker’s melanosis were observed during oral examination. In addition, white lesions interspersed with the reddish ones were located bilaterally in the retrocomissural region. The lesions had a triangular shape and were painless. Clinically, the lesion on the right side was more apparent during the examination (Figure 1).

Clinically, the lesion was a plate with approximate size of 20 mm, and white color interspersed with red areas. It had a slightly rough consistency with sessile insertion and absence of symptoms.

The diagnosis of chronic hyperplasic candidiasis was considered initially because of the clinical appearance of the lesions, and smoking and drinking habits. The patient was not aware of the existence of lesions. Therefore, it was not possible to establish the time of lesion development. During medical interview, the patient reported that he had drunk alcohol every day. In addition, he had smoked on average two packs of cigarettes a day for over 20 years.

Before dental treatment, the patient underwent an incisional biopsy of the lesion. However, it was decided to completely remove the lesions because of their location and size. The diagnostic hypotheses were chronic hyperplasic candidiasis or erythroleukoplakia.

The patient was medicated with 200 mg of Ibuprofen in a therapeutic regimen of doses administered every 12 hours for 3 days. Seven days later, he returned for suture removal. No change was observed during the repair process.

The lesions were sent for histopathological examination. The material was processed in the laboratory and stained by two stains:
hematoxylin and eosin and Grocott. The hematoxylin and eosin revealed a fragment of oral mucosa lined by parakeratinized epithelial tissue with areas of hyperplasia, acanthosis, and exocytosis. Some cells of the basal layer exhibited enlarged nuclei with condensed chromatin, nuclear atypia and mitotic figures (Figures 2 and 3). The underlying connective tissue showed chronic inflammatory infiltrate with a predominance of lymphocytes. No fungal structures were identified with Grocott staining (Figure 4). That finding excluded the possibility of fungal infection of the lesion. According to those histological findings, the diagnosis of mild epithelial dysplasia was established.

The patient was advised to stop or reduce the consumption of cigarettes and alcohol because of the diagnosis of epithelial dysplasia and nicotine stomatitis. Periodic examinations were planned but the patient did not return for the follow-up.

DISCUSSION

Oral leukoplakia is defined as a white plate which cannot be clinically or histologically characterized as any other disease [3]. Thus, leukoplakia is essentially a clinical entity. Its diagnosis is based on exclusion of other injuries, because this entity has no specific histopathological features [14]. It is considered the most common potentially malignant disorder that develops in several areas of the oral mucosa. However, the buccal mucosa, lower lip and edge of the tongue are the most affected areas. Erythroplakia is defined as no red plate diagnosed histologically or any other condition [8]. This lesion has a high degree of malignancy compared with oral leukoplakia or nevus [15].

When the lesion presents with red and white mucosal alterations concomitantly, the term erythroleukoplakia is used. However, in erythroleukoplakia lesions, the red or erythroplakia areas have been shown to be most likely to demonstrate dysplastic changes compared to the white hyperkeratotic areas [16].

It is believed that the etiology of this lesion is related to habits such as smoking and when associated with alcohol consumption, the probability of malignant transformation is increased. They are generally considered idiopathic lesions and without apparent origin. The erythroleukoplakias are more prevalent in men, due to an increased association with smoking [17].

A study by Feller et al. [18] revealed that the prevalence of erythroleukoplakias is approximately 11.2%. The same study observed that it mainly affects males and individuals in their seventh decade of life. In our case report, the patient was male, 37 years of age, smoked cigarettes (40 cigarettes a day) and drank alcohol (daily). The anatomical regions most affected by potentially malignant...
disorders are: buccal mucosa (28.8%), floor of the mouth (18.3%), alveolar ridge and gums (17.3%), and tongue (12.0%). The patient reported here had a lesion within the buccal mucosa.

The initial diagnosis of this case was chronic hyperplastic candidiasis due to the clinical aspect of the lesion and patient's habits of cigarette smoking and alcohol drinking. Some white lesions need to be considered in the differential diagnosis of oral potentially malignant disorders with white coloration, i.e.: pseudomembranous and chronic hyperplastic candidiasis, linea alba, lichen planus, leukoedema, reactive hyperkeratosis due to oral mordiscamento and spongy white nevus [8]. Initially, pseudomembranous candidiasis was excluded because it was not possible to remove the lesion with scraping. In addition, the lesions of the patient had no clinical features consistent with the diagnosis of other possibilities mentioned above. The only exception was reaction hyperkeratosis due to cheek bite. However, the patient reported no parafunctional habit, nor was there any evidence found during the clinical examination. Thus, one was not able to rule out the possibility of a bite injury in the same region during sleep, since the patient complained of pain in the TMJ region.

As chronic hyperplastic candidiasis was the first diagnostic hypothesis, we opted to perform an incisional biopsy. However, during surgery it was decided to completely remove the lesions into cancerous ones, the combination of clinical and histopathological examination is critical for accurate diagnosis, and the results from these assessments are used to determine specific treatment [20]. Thus, dentists and otolaryngologists need to be alert during the examination of the mouth to the habit of smoking and drinking of the patient. Oral biopsy is mandatory to recognize the presence and the severity of epithelial dysplasia, which is a decisive factor for subsequent treatment planning.

The histopathological findings that characterize leukoplakia are hyperkeratosis, dysplasia, chronic inflammatory infiltrate, acanthosis, and atrophy. According to the classification of dysplasia (mild, moderate and severe), two lesions with severe dysplasia are already considered carcinoma in situ [8]. However, histopathological examination revealed that this lesion presented hyperkeratosis, acanthosis, atrophy, mild dysplasia, and chronic inflammatory infiltrate. In addition, staining by Grocott ruled out the possibility of chronic hyperplastic candidiasis.

Erythroleukoplakia can be treated by surgical removal at the time of the biopsy, or surgical laser, photodynamic therapy, and chemopreventive agents [11,12,13,19]. Thus, it is necessary that the diagnosis is made in advance and safely. In cases where moderate to severe dysplasia is evident, complete surgical removal and periodic monitoring are recommended due to high rates of recurrence [8].

The importance of early diagnosis of erythroleukoplakia follows from a potential for dysplastic changes and progression to frank carcinoma. Because of the rate and timing of progression of these lesions into cancerous ones, the combination of clinical and histological examination is critical for accurate diagnosis, and the results from these assessments are used to determine specific treatment [20]. Thus, dentists and otolaryngologists need to be alert during the examination of the mouth to the habit of smoking and drinking of the patient. Oral biopsy is mandatory to recognize the presence and the severity of epithelial dysplasia, which is a decisive factor for subsequent treatment planning.

References


