BILATERAL RECURRENT SPECKLED LEUKOPLAKIA: A CASE REPORT

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ABSTRACT:

Largely oral cancers are preceded by potentially malignant lesions, which may appear as white or red patches on the oral mucosa. Leukoplakia is one of the most common epithelial precursors of oral squamous cell carcinoma. Speckled leukoplakia is a rare type of leukoplakia with a very high risk of premalignant growth and mortality rate. Though it is the common precancerous lesion, it poses a major diagnostic and therapeutic challenge. We present a rare case of bilateral recurrent speckled leukoplakia with malignant transformation and discuss this relatively rare entity in light of current information from the literature. We also attempt to present the clinical relevance, and the therapeutic modalities available for the management of the disease.

Key words: Speckled Leukoplakia, White Patch, Tobacco Chewing, Precancerous Lesions

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INTRODUCTION:
Oral leukoplakia is characterized by adherent white plaques or patches on the mucous membranes of the oral cavity, including the tongue [1, 2]. The World Health Organization (WHO) currently employs the term Speckled Leukoplakia (SL) to describe the presence of both white and red patches on the oral mucosa [3, 4]. SL is a rare, highly aggressive, clinicopathological entity, with high-risk of malignant transformation and a precursor lesion of squamous cell carcinoma [2, 3]. Although the prevalence of SL in India is low, it presents histopathological features ranging from epithelial dysplasia to invasive carcinoma. This justifies placing these lesions among the oral lesions with the highest malignant potential.

CASE REPORT:
A 58-year-old male patient reported to the department of oral medicine and radiodiagnosis, with the complaint of painless white patch on right and left buccal mucosa since 4 months. Patient had a habit of cigarette smoking, 5 to 7 per day for a period of 30 years and presently patient had reduced smoking to 3 per day. He also had a habit of chewing unprocessed tobacco since 10 years with a frequency of 2 to 3 times a day. The content of the quid was betel leaf, areca nut, slaked lime, and tobacco. Along with these the patient also consumed alcohol occasionally.

Intraoral Examination of right and left buccal mucosa revealed firm, non-tender, non-scrapable, red and white patches measuring 2 x 2 cm. Surface appears rough and slightly elevated which clinically resembled “mud crack” in appearance. The lesion was not disappearing on stretching (Fig. 1). Chair side investigation, toluidine blue staining was carried out to select the area of biopsy to be made. The selected area was then biopsied (incision).

Histopathologic examination with H and E stained sections showed hyperkeratotic stratified squamous epithelium overlying a fibrous connective tissue. Epithelium was showing hyperplasia with moderate to severe dysplastic features. Dense inflammatory infiltrate were also seen. The overall clinical and histopathological findings were considered diagnostic for SL with moderate to severe dysplasia. The patient underwent complete surgical removal of the lesion along with silicon graft (Fig 2). Post operative healing was uneventful (Fig 3). Patient was asked to visit the hospital for a regular check up once in a month.
Fig. 1: Red and white patch on right buccal mucosa

Fig. 2: Surgical excision on right buccal mucosa

Fig. 3: Healed surgical site after 3 weeks of surgery

Fig. 4: Recurrence of speckled leukoplakia after 3 months

Fig. 5: Histopathological photograph showing moderate to severe dysplasia
On 6 months follow up a recurrent lesion on the same buccal mucosa were seen. The lesion on the buccal mucosa was now showing more red areas on the surface (Fig. 4). They were diagnosed as SL. Both the lesions were taken for excisional biopsy. The H and E stained sections of both the lesions showed moderate to severe dysplastic features (Fig. 5). The patient is still under periodic follow-up once in every three months.

**DISCUSSION:**

WHO defines leukoplakia as a whitish patch or plaque that cannot be characterized, clinically or pathologically, as any other disease and which is not associated with any other physical or chemical causative agent except the use of tobbacco [5]. The literature, however, strongly indicates the role of alcohol, viruses and systemic conditions [6, 7].

Various designations have been used to describe the presence of both white and red patches. Lesions appearing completely red are named as Erythroplakia. SL is indicated when red and white patches are present over the mucosa [2, 3]. WHO [4] currently recommends the term SL to describe mouth lesions that present red and white components; hence this term is used in the present case report.

Men are more commonly affected and the mean age at the time of diagnosis is slightly over 60 years [8, 9, and 10]. The buccal mucosa and tongue are the most common sites associated with SL; palatal mucosa, alveolar mucosa, gingiva, floor of mouth, and lip show a lower incidence. Typically, multiple oral sites can be also affected [11, 12], as seen in the present case.

Our case highlights the typical slow-growing, progressive, and persistent clinical course of this rare condition. As presented in our case, the initial finding can be an elevated non homogeneous whitish-grey lesion that tends to recur and proliferate, often over a protracted period of time, to result in a diffuse and widespread erythematous plaque that was associated with severe dysplasia.

Non-homogeneous leukoplakias are often associated with mild complaints of localized pain or discomfort and invariably develop into malignancy. Approximately 80 percent of SL progress to oral carcinoma over a period of time in spite of a variety of interventions. This feature contrasts with homogenous leukoplakia in which approximately 5-10 percent will transform into a carcinoma [2]. SL is resistant to most of the available treatment modalities, including surgery [4]. Therefore, total excision with free surgical margins is critical combined with a lifelong follow-up [4].

Malignant potential of leukoplakia is higher in women (6%) than in men (3.9%). Leukoplakia associated with habit of chewing tobacco shows higher rate of malignant transformation as compared to others [15]. In buccal mucosa and commissure region 1.8 percent malignant
transformation can occur. In lip and tongue region 16 to 38.8 percent malignant transformation has been reported. The annual malignant transformation rate has been determined to be 0.1% to 17% [16]. Less than half (33% to 42%) of leukoplakias undergo malignant change [17].

**MANAGEMENT:**
The degree of epithelial dysplasia plays a pivotal role while deciding the nature of treatment to the patient. Martorell-Calatayud [18] defined two risk groups and the subsequent treatment options:

**Group 1:** Those with low risk of malignisation:
Those leukoplakias lacking dysplasia, and those that show mild dysplasia located in low risk areas or those with a thickness of less than 200 mm or that present clinically as homogenous leukoplakia. A range of therapeutic approaches can be taken in this group:
- Regular patient follow-up. The interval between follow-up visits should not exceed 12 months in order to detect any change, suggestive of malignant transformation.
- Treatment of lesions with topical or oral retinoids {eg: 13-Cis-Retinoic Acid (1.5 to 2 mg/kg body weight for 3 months)} [18].
- Treatments using nonsurgical ablative techniques, such as cryotherapy and carbon dioxide laser ablation. Of these options, the use of laser light has shown better results in terms of controlling the lesions, and so it is considered the treatment of choice in this low risk group.

**Group 2:** Those with high-risk of malignant transformation, which comprises:
- Those leukoplakias with mild dysplasia located in high-risk areas measuring more than 200 mm, or those associated with a nonhomogenous clinical form; Leukoplakias with moderate or severe dysplasia; Verrucous leukoplakias. In this group, aggressive surgical treatment, consisting of excision of the entire thickness of the mucosa at the site of the leukoplakia is recommended. This is similar to the present case.
- Among the many therapeutic options available, however, eliminating risk factors (smoking, alcohol) and etiological factors (sharp broken teeth, faulty metal restorations and metal bridges) are preventive measure applicable to all patients with these lesions [13]. Regular check-up of these patients is essential, probably every 3, 6 and then 12 months, both in treated and untreated patients.

**CONCLUSION:**
Dentists, Dermatologists, ENT specialists, or Physicians may encounter asymptomatic initial and progressive lesions of leukoplakia on routine clinical examination. Patients with SL are benefitted if we have the earliest possible diagnosis and treatment, thus improving their prognosis. All the patients with a recurrent
white lesion require vigorous follow up and have to be treated with aggressive approach.

REFERENCES: