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Case Report Open Access

Verrucous squamous cell carcinoma arising in oral submucous fibrosis, An Infrequent Overlap of Dual Morbidities-case report

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Abstract

Background

Oral submucous fibrosis is a chronic, progressive potentially malignant disorder commonly associated with areca nut and tobacco consumption. The condition is characterized by mucosal fibrosis, restricted mouth opening, and a significant risk of malignant transformation. Although the most frequent malignancy arising in this background is conventional squamous cell carcinoma, verrucous squamous cell carcinoma represents a distinct low-grade, well-differentiated variant with characteristic clinical and histopathological features.

Case Presentation

A 37-year-old male presented with a history of burning sensation on consumption of spicy food and progressive restriction in mouth opening for six months, along with a gradually enlarging growth on the right buccal mucosa. Clinical examination revealed features suggestive of oral submucous fibrosis, including diffuse mucosal blanching, fibrotic bands, shrunken uvula, and reduced mouth opening measuring 21 mm. An exophytic verrucous growth was observed on the right buccal mucosa extending from the commissure to the molar region. The patient underwent wide local excision of the lesion with supraomohyoid neck dissection followed by reconstruction using a radial forearm free flap. Histopathological examination of the excised specimen revealed well-differentiated verrucous squamous cell carcinoma with clear surgical margins and no evidence of regional lymph node metastasis (pT1N0).

Conclusion

This case highlights the occurrence of verrucous squamous cell carcinoma arising in a background of oral submucous fibrosis and emphasizes the importance of early clinical recognition, thorough evaluation, and definitive surgical management. Regular follow-up of patients with oral submucous fibrosis is essential for early detection of malignant transformation and improved clinical outcomes.

Keywords: Oral submucous fibrosis; verrucous squamous cell carcinoma

Introduction

Oral submucous fibrosis (OSMF) is a chronic, progressive, and potentially malignant disorder of the oral cavity characterized by juxta-epithelial inflammatory reaction and progressive fibrosis of the lamina propria and deeper connective tissues, leading to stiffness of the oral mucosa and limitation of mouth opening. The condition is predominantly reported in South and Southeast Asian populations and is strongly associated with the habitual use of areca nut, betel quid, and tobacco products [1, 4]. Clinically, OSMF presents with burning sensation of the oral mucosa, blanching, formation of fibrotic bands, intolerance to spicy food, and progressive trismus [2, 3]

The pathogenesis of OSMF is multifactorial, with areca nut alkaloids such as arecoline playing a significant role in stimulating fibroblast proliferation and increased collagen synthesis, resulting in excessive deposition of collagen in the oral mucosa. Additional contributing factors include chronic inflammation, nutritional deficiencies, and genetic susceptibility [2, 4].

OSMF is widely recognized as a potentially malignant disorder with a reported malignant transformation rate ranging from approximately 7% to 13%. The most common malignancy arising in this background is conventional squamous cell carcinoma. However, less common histopathological variants, including verrucous carcinoma, may also develop in patients with long-standing OSMF [3, 5, 11].

Verrucous carcinoma is a well-differentiated, low-grade variant of squamous cell carcinoma first described by Ackerman in 1948 [6]. It is characterized clinically by a slow-growing exophytic lesion with a verrucous or cauliflower-like surface and histologically by hyperkeratotic epithelium with papillomatous projections, broad bulbous rete ridges, and minimal cytological atypia [6, 8]. This tumor typically demonstrates locally aggressive growth with a pushing pattern of invasion but has a very low potential for regional or distant metastasis [7, 9].

Several studies have described the clinicopathological features of verrucous carcinoma of the oral cavity and emphasized the importance of early diagnosis and surgical management to achieve favorable outcomes [9, 12, 13]. The occurrence of verrucous squamous cell carcinoma in association with oral submucous fibrosis represents an important clinicopathological entity because the fibrotic changes and restricted mouth opening may delay clinical detection and management.

The present report describes a case of verrucous squamous cell carcinoma arising in a background of oral submucous fibrosis in a young male patient with a long history of areca nut and tobacco chewing, highlighting the clinical presentation, histopathological features, and surgical management of this uncommon association.

Case report

A 37-year-old male patient presented with a chief complaint of burning sensation in the oral cavity while consuming spicy food and progressive difficulty in mouth opening for the past six months. The patient also reported the presence of a growth on the inner aspect of the right cheek that had gradually increased in size over the same duration. The lesion was not associated with pain or bleeding but caused mild discomfort while eating.

The patient gave a history of habitual tobacco and betel nut chewing for approximately 10 years with a frequency of multiple times per day. There was no relevant medical or surgical history.

On extraoral examination, no obvious facial asymmetry was noted. Mild tenderness in the preauricular region was observed during mouth opening. Single right submandibular lymph nodes palpable, which is freely movable not attached to the underlying structures

Intraoral examination revealed diffuse blanching of the oral mucosa involving the bilateral buccal mucosa, palate, and extending to the floor of the mouth. The uvula appeared shrunken and fibrosed. Multiple inflamed minor salivary gland openings were noted on the lower labial mucosa and soft palate. Vertical fibrotic bands were palpable bilaterally in the buccal mucosa extending posteriorly to the retromolar region. Circumoral fibrotic bands were also noted around the lips, which appeared shrunken and rubbery in consistency. The interincisal mouth opening was approximately 21 mm and the patient reported severe burning sensation with a visual analog scale score of 10.

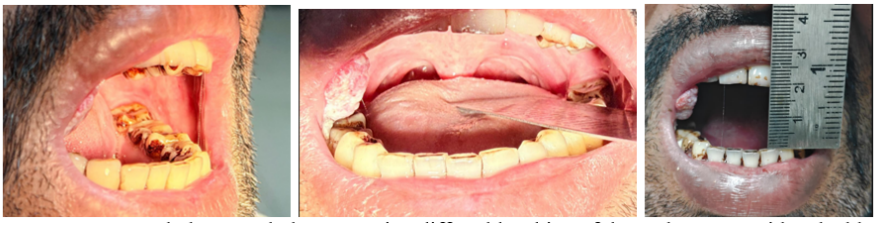

Figure 1: Intraoral photograph demonstrating diffuse blanching of the oral mucosa with palpable fibrotic bands and restricted mouth opening

A solitary exophytic proliferative lesion was present on the right buccal mucosa extending from the commissure of the lip posteriorly up to the region of the lower second molar and vertically from the maxillary vestibule to the mandibular vestibule. The lesion appeared irregular with a verrucous surface and whitish keratotic areas. On palpation, the lesion was firm in consistency, non-tender, and exhibited a broad sessile base.

Figure 2: Intraoral photograph showing an exophytic verrucous proliferative lesion on the right buccal mucosa.

Based on the clinical findings, a provisional diagnosis of oral submucous fibrosis with a verrucous carcinoma of the right buccal mucosa was considered.An incisional biopsy was performed from the lesion, and the initial histopathological examination suggested verrucous hyperplasia. Considering the clinical suspicion and the proliferative nature of the lesion, the patient was planned for definitive surgical management.

The patient subsequently underwent wide local excision of the lesion involving the right buccal mucosa with supraomohyoid neck dissection. Reconstruction of the surgical defect was performed using a radial artery forearm free flap. Histopathological examination of the excised specimen revealed verruciform epithelial proliferation with marked hyperkeratosis, lamellated orthokeratosis and parakeratosis, broad bulbous rete ridges extending into the underlying connective tissue with pushing margins, and minimal cytological atypia. Prominent keratin clefts and keratin pearls were also observed. These findings were consistent with a diagnosis of well-differentiated verrucous squamous cell carcinoma.

The tumor measured approximately 2 × 1.6 × 0.9 cm. All surgical margins were free of tumor, with the closest margin measuring 0.4 cm. Examination of the excised lymph nodes revealed no evidence of metastatic involvement. The tumor was staged as pT1N0 according to the AJCC staging system.

The postoperative period was uneventful, and the patient recovered well following surgery. The patient was advised regular follow-up and cessation of tobacco and betel nut chewing habits.

Discussion

Oral submucous fibrosis (OSMF) is a chronic, progressive, and potentially malignant disorder characterized by fibrosis of the oral mucosa, resulting in stiffness of the oral tissues and progressive limitation of mouth opening. The condition is strongly associated with the habitual use of areca nut and tobacco products and is commonly reported in South and Southeast Asian populations.[1–4] Due to continuous exposure of the oral mucosa to carcinogenic substances present in these products, patients with OSMF carry a significant risk of malignant transformation. The reported transformation rate ranges from approximately 7% to 13% [3, 5].

Although malignant transformation in OSMF is well documented, the occurrence of verrucous carcinoma in association with OSMF is considered relatively uncommon. Verrucous carcinoma is a distinct clinicopathological entity first described by Ackerman in 1948 and is characterized by a slow-growing exophytic lesion with a verrucous or cauliflower-like surface [6]. Clinically, these lesions present as well-demarcated proliferative growths with a thick keratinized surface and broad base [7].

Histopathologically, verrucous carcinoma demonstrates characteristic features including verruciform epithelial proliferation, marked hyperkeratosis, and broad bulbous rete ridges extending into the underlying connective tissue with pushing margins. Cytological atypia is minimal, and the tumor generally shows a locally aggressive growth pattern with a low tendency for regional or distant metastasis [7,9]. These distinctive features help differentiate it from other proliferative lesions of the oral cavity.

The coexistence of OSMF and veracious carcinoma has been reported only in a limited number of studies. The chronic irritation caused by areca nut and tobacco, which are major etiological factors for OSMF, may also contribute to the development of verrucous carcinoma. Continuous exposure to these carcinogens results in epithelial alterations that may predispose the mucosa to neoplastic changes [2,4,12].

In the present case, the patient exhibited classical clinical features of OSMF including diffuse mucosal blanching, palpable fibrotic bands, shrunken uvula, and restricted mouth opening measuring 21 mm. Additionally, a proliferative verrucous lesion was observed on the right buccal mucosa, which is one of the commonly affected sites for verrucous carcinoma in individuals

with long-standing habits of tobacco and areca nut chewing [9]. The frequent placement of the quid in the buccal vestibule exposes the mucosa to chronic irritation and carcinogenic substances, which may contribute to the development of such lesions.

Histopathological examination of the excised specimen in this case revealed classical features of verrucous carcinoma, including verruci form epithelial proliferation, hyperkeratosis, and broad rete ridges with pushing margins. These findings are consistent with previously reported descriptions of verrucous carcinoma in the literature [6,8].

Surgical excision with adequate margins is considered the treatment of choice for verrucous carcinoma and generally results in favorable outcomes due to the low metastatic potential of this tumor [9,13].The present case further highlights the importance of careful clinical evaluation and regular follow-up in patients with OSMF, as early detection of suspicious proliferative lesions allows timely diagnosis and appropriate management.

Conclusion

Oral submucous fibrosis is a potentially malignant disorder that requires careful clinical evaluation and long-term follow-up due to its risk of malignant transformation. Although the development of verrucous carcinoma in the background of oral submucous fibrosis is relatively uncommon, clinicians should maintain a high index of suspicion when evaluating proliferative lesions in patients with long-standing areca nut and tobacco habits. Early recognition through thorough clinical examination, appropriate histopathological assessment, and timely surgical management plays a crucial role in achieving favorable outcomes. The present case highlights the importance of comprehensive evaluation of suspicious lesions in patients with oral submucous fibrosis to facilitate early diagnosis and effective management of such rare occurrences.

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