Spindle cell carcinoma of the gingiva: A rare occurrence

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Abstract

Of the many neoplasms known, squamous cell carcinoma (SCC) is the most common to affect the oral cavity. Spindle cell carcinoma (SpCC) is considered a rare high malignant variant of SCC occurring predominantly in the upper aerodigestive tract. Soft-tissue spindle cell neoplasms are quite uncommon in the oral cavity reportedly accounting for lesser than 1% of all tumors in the oral region. Our case shows an unusual presentation of SpCC involving the mandibular gingiva in a 46-year-old smoker patient, which presented as a firm, erythematous swelling with surface necrosis. An incisional biopsy was performed for microscopic evaluation to confirm the clinical diagnosis and for treatment planning.

Keywords: Gingiva, smoking, spindle cell carcinoma

Introduction

Squamous cell carcinoma (SCC) is by far the most important and common malignant mucosal neoplasm to affect the head and neck region. Occasionally, variants of SCC may be encountered, which includes verrucous, exophytic or papillary, spindle cell (sarcomatoid), basaloid and adenosquamous types that make up in aggregate for about 10-15% of all SCCs.^[1]

Spindle cell carcinoma (SpCC) also called Lane tumor is an uncommon poorly differentiated type of SCC comprising up to 3% of SCC.^[1] SpCC has been referred to by a variety of names such as pseudosarcoma,^[2] carcinosarcoma^[3] and pleomorphic carcinoma, which reflects the divergent interpretations of the sarcomatoid component as reactive or neoplastic and mesenchymal or epithelial. There is a profound male to female predilection (11:1) and generally occurs in individuals in their seventh decade of life. According to Ellis,^[4] the frequent sites of occurrence of SpCC are the lower lip, tongue and alveolar ridge. Gingival involvement seems to be a rarity.^[5,6]

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This report describes a rare presentation of SpCC of the gingiva with a low metastatic potential contrary to the aggressive nature of the tumor.

Case Report

A 46-year-old male patient reported to the Department of Periodontology, Faculty of Dental Sciences, Sri Ramachandra University, Chennai, India with an intraoral swelling in relation to the lower left first and second premolars for the past 8 months associated with a dull pain. The swelling was initially small and gradually increased in size. He was a known asthmatic, but not under any systemic medication. He gave a history of smoking cigarettes (2 packets/day) for the past 30 years and keeping the tobacco quid on the left side of the vestibular area, but had stopped the habit because of burning sensation. On clinical examination, a single sessile erythematous swelling with a fungating whitish mass in the center was seen [Figure 1]. On palpation, it was firm in consistency measuring about 1.5 cm (apicocoronally) and 2.0 cm (mesiodistally) extending from the mesial aspect of tooth 34 to the mesial aspect of tooth 36. Both the premolars exhibited grade 1 mobility with tenderness on vertical percussion and no attachment loss. A single submandibular lymph node was palpable on the left side.

Patient was counseled regarding the cessation of the smoking habit. Scaling was performed on the first visit and selective grinding of the opposing cusp in occlusion was carried out. At the recall visit, the surface necrosis had disappeared (which could have probably been due to withdrawal of tobacco quid usage).

Complete hemogram, intraoral periapical radiograph and orthopantomograph were taken. Except for an increase in the eosinophil count, all the other blood parameters were within normal range. No significant alterations were evident in the radiographs [Figure 2]. Based on the clinical findings, a provisional diagnosis of SCC was given. An incisional biopsy of the growth was performed under local anesthesia. The tissue specimen was immediately transferred into 10% buffered formalin solution, sent for routine histopathological examination and were further subjected to immunohistochemical (IHC) analysis.

Histologically, the tumor showed an ulcerated parakeratotic stratified squamous epithelium [Figure 3] with a focus of epithelial pearl formation [Figure 4]. The underlying connective tissue stroma revealed richly cellular pleomorphic spindle cells arranged in fascicles resembling a herring bone pattern characteristic of a fibrosarcoma [Figure 5]. The sections were further subjected to IHC for cytokeratin, vimentin and CD34 expression. IHC analysis revealed spindle shaped cells that showed positive reactions for cytokeratin [Figure 6] and vimentin [Figure 7] but negative for CD34 [Figure 8]. Antibody to podoplanin, a molecule expressed in lymphatic endothelial cells was used. Positive membrane immunoreactivity was observed in the basal cell layer and connective tissue stroma with a relatively increased lymphatic network complexity as compared with the tumor free zone [Figure 9].

A whole body plain and contrast computed tomography (CT) view was performed. CT view of mandible with contrast revealed a 17.8 mm \times 4.7 mm sized soft-tissue density lesion in the buccal aspect of the body of the mandible on the left side with no evidence of bony erosion [Figure 10]. Few sub centimeter sized lymph nodes were present in the left submandibular [Figure 11] and bilateral deep cervical (level II) regions [Figure 12].

Patient was referred to an oncologist. Following chemotherapy, the patient was subjected to surgical management that involved a segmental resection of the mandible with supraomohyoid neck dissection followed by radiation therapy.

Discussion

SpCC is a rare type of SCC that most often involves the larynx and gingiva is a rare site for its occurrence. In the present case, the placement of the tobacco quid in that region could have contributed to this unusual site predilection since the activity of the carcinogens is generated through deoxyribonucleic acid adducts.^[7] SCC most commonly affects men than women, usually in the middle or later decades of life, (although any age can be affected). In accordance, this lesion reported was present in a 46-year-old male patient. Clinical presentation of oral SpCC can vary from an exophytic polypoid mass with an ulcerated surface to a frankly infiltrative ulcer, occurring mainly in the alveolar ridge.^[8] In this case, SpCC presented clinically as a polypoid growth with surface necrosis involving an unusual location, i.e., the mandibular gingiva.

Although there are various risk factors such as age, genetic, viruses, environment and occupation, which can influence

the occurrence of SpCC, four predominant factors, which are considered to predispose for the development of SpCC are alcohol abuse, poor oral health, previous irradiation to the area of the tumor and the most important risk factor being tobacco in various forms such as cigarette, cigar, pipe and smokeless tobacco.^[8]



Figure 1: Initial appearance of the lesion with a fungating whitish mass on the surface



Figure 2: Orthopantomograph revealing no hard tissue abnormalities



Figure 3: Photomicrograph of spindle cell carcinoma showing stratified squamous epithelium with surface ulcerations, granulation tissue and mixed inflammatory cells (H and $E, \times 10$)



Figure 4: Photomicrograph showing epithelial pearl formation (arrow) (H and E, ×40)



Figure 5: Photomicrograph of spindle cell carcinoma showing richly cellular pleomorphic, hyperchromatic spindle cells arranged in fascicles depicting a "herring bone" pattern (arrow) (H and E, ×10)



Figure 6: Spindle shaped tumor cell component immunostained with anti-human cytokeratin antibody showing positive immunostaining for cytokeratin (×40)



Figure 7: Spindle shaped tumor cell component immunostained with anti-human vimentin antibody showing positive immunostaining for vimentin (×40)



Figure 8: Spindle shaped tumor cell component immunostained with anti-human CD34 antibody. This component shows negative immunostaining of spindle cells for CD34 and positive immunostaining of endothelial cells (×40)

SpCC is considered to be basically aggressive in nature because the incidence of metastases was found to be 36% and the 2-year survival rate was 55% in tumors involving



Figure 9: Positive expression of podoplanin in the spindle cells and increased lymphatic network complexity (×40)

the oral cavity.^[6] Our present case indeed was unique since the tumor was slowly progressive over 8 months duration with no signs of distant metastases contrary to the reported aggressive nature of conventional SpCC.^[9,10]



Figure 10: Computed tomography view of mandible with contrast depicting a soft tissue density lesion in the buccal aspect of the body of the mandible on the left side (arrow)



Figure 12: Computed tomography view of the mandible with contrast depicting few sub centimeter sized lymph nodes in the bilateral deep cervical region (arrows)

In SpCC, the pleomorphic component originates through dedifferentiation of the SCC component. The most sensitive and reliable epithelial marker cytokeratin was used for demonstration of the epithelial phenotype. Positive cytokeratin and vimentin expression was seen, but the tumor cells were negative for CD34 (only the endothelial cells showed a positive reaction) eliminating the possibility of any vascular malignant tumors. Podoplanin, a molecule expressed in malignant cells was used to determine for any lymphatic invasion. To the best our knowledge, this is one of the first cases to have employed a lymphatic marker for SpCC of the gingiva.

Conclusion

To conclude, SpCC can occur on the gingiva and since exposure to these tumors is infrequent for clinicians, it



Figure 11: Computed tomography view of the mandible with contrast depicting few sub centimeter sized lymph nodes in the left submandibular region (arrow)

is imperative to reiterate that all patients have an early diagnostic biopsy before the definitive therapy for a better prognosis and thereby reducing morbidity and mortality.

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