

Spindle Cell Squamous Cell Carcinoma of the Tongue: A Rare Variant at an Even Rarer Location

Abstract

Spindle cell carcinoma (SpCC) is an unusual variant of squamous cell carcinoma (S.C.C.) of the head and neck region. It is a rare biphasic neoplasm. SpCC is an unusual morphological variant of S.C.C. It is characterized by the presence of both carcinomatous as well as sarcomatous component and accounts for 2 to 3% of all the S.C.C.s. Seven years ago, the W.H.O. classification has placed this tumor under the malignant epithelial tumors and called it SpCC. The histogenesis of spindle cells remains controversial and is believed to be monoclonal epithelial neoplasia with a close association with squamous carcinoma cells. Since SpCC is a rare tumor, its histopathological diagnosis is often very complex. Immunohistochemistry (I.H.C.) supports the epithelial nature of this tumor. Both neoplasia components possess immunoreactivity for cytokeratin and vimentin. We are presenting a case of a middle-aged man who was diagnosed with this rare variant at an even rarer location.

Keywords: *Biphasic tumour, Head & Neck, Spindle cell carcinoma, Squamous cell carcinoma, Tongue*

Introduction

The tongue's spindle cell carcinoma (SpCC) is a comparatively rare neoplasm of the head and neck region. It is a biphasic neoplasm. It is described as an unusual morphological variant of S.C.C. It was first described by Virchow in 1865.^[1] It is characterized by the presence of both carcinomatous as well as sarcomatous elements and accounts for 2 to 3% of all S.C.C.s in the head and neck region.^[2-4] It is an aggressive tumor showing local recurrence and metastasis.^[5] It principally occurs in the upper digestive tract, more commonly in the larynx and other sites like the nasal cavity, hypopharynx, esophagus, trachea, breast, oral mucosa, and much more rarely in the tongue.^[6, 7] Most cases show male preponderance and are most commonly seen between the sixth and seventh decade of life. It is seen to be associated with alcohol use, smoking, and radiation exposure in the affected areas. SpCC usually presents as a large pedunculated and polypoidal mass with focal surface ulceration.^[2, 4, 7]

Since SpCC is a rare tumor, its histopathological diagnosis is often very complex. The histogenesis of spindle cells

remains controversial and is believed to be a monoclonal epithelial neoplasia with a close association with squamous carcinoma cells. A single stem cell gives origin to both the cell components wherein further differentiation or metaplastic alteration leads to a spindle cell formation.^[8] The I.H.C. supports the epithelial nature of the neoplasm as both neoplasia components possess immunoreactivity for cytokeratin, vimentin, and epithelial membrane antigen (E.M.A.) and the do not show immunoreactivity for S-100 or smooth muscle actin alpha.^[9, 10] The prognosis of this carcinoma is not favorable.^[7]

Case report

A 35-year-old man, a tobacco chewer and a heavy alcohol drinker, came to the hospital with complaints of difficulty in speaking and significant weight loss for six months. The patient had a previous history of carcinoma of the tongue five years back; for which the patient was subjected to wide local excision. Physical examination revealed a 3 cm × 2 cm ulceroproliferative growth on the left lateral border of the tongue, along with enlarged bilateral cervical lymph nodes. FDG PET CT scan reported a heterogeneously enhancing soft tissue density ulcerative lesion along the left

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lateral border of the tongue.

Gross examination revealed a unifocal 3.0cm × 2.0 cm × 1.0 cm, grey-white, hard ulcero-proliferative growth, which was 0.8 cm away from closest margin (base). The cut section revealed a greyish-white, firm surface with a variegated appearance and few areas of ulceration or hemorrhage without necrosis (**Figure 1a**). Microscopic examination showed extensive areas of granulation tissue along with sub-epithelium showing two neoplastic cellular components, one with the proliferation of oval and elongated, pleomorphic spindle cells arranged in short and long fascicles and the second with nests of squamous epithelial cells with marked

pleomorphism, cellular atypia, scanty cytoplasm, irregular and hyperchromatic nuclei, coarsely clumped chromatin, and one to two prominent nucleoli along with foci of atypical mitosis. Lymphovascular and perineural invasion was not identified (**Figures 1b and 1c**). All margins were free of tumors. On I.H.C., the tumor cells were immunopositive for Cytokeratin (**Figure 1d**), vimentin and p40 (focal nuclear positive). The markers CD34 and Smooth Muscle Actin alpha remained negative. Ten lymph nodes were identified in the modified neck dissection, and all were free from the tumor. The final histopathological diagnosis was given as a spindle cell variant of Squamous Cell Carcinoma (SpCC) with stage rpT3N0Mx grade 3. The patient was advised for neo-adjuvant radiotherapy and is currently on follow-up.

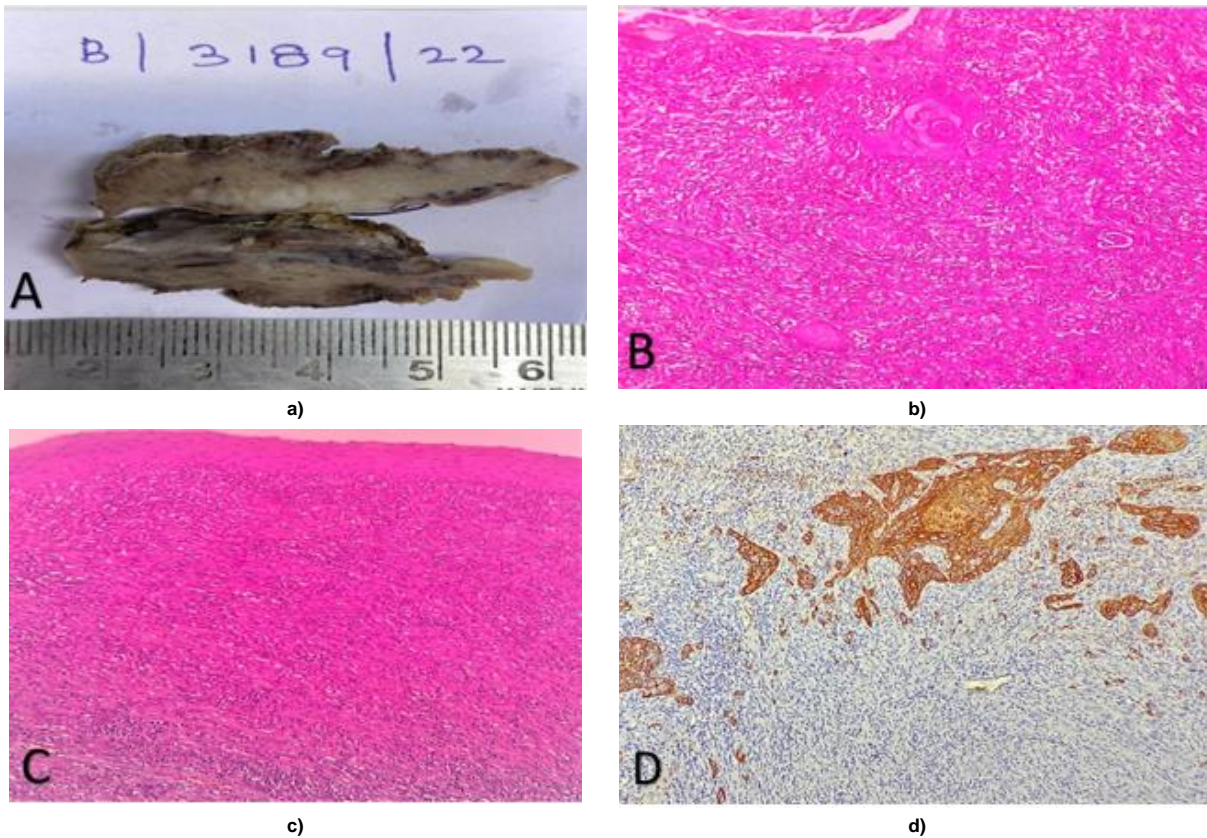


Figure 1. a) Shows grey-white cut section of ulceroproliferative growth of tongue. b) Photomicrograph showing Squamous epithelial cells component of the tumor. c) Photomicrograph showing Spindle cells component of the tumor. d) Immunohistochemical stain CK 5/6 shows positive staining for the squamous epithelial cell component and negative staining for the spindle cell component.

Results and Discussion

SpCC is a rare neoplasm accounting for 2-3% of S.C.C.s of the head and neck region. These tumors are biphasic, comprising both carcinomatous and sarcomatous components. Very limited literature is available regarding its localization on the tongue at such a young age. Many terms, including sarcomatoid carcinoma, pseudo sarcoma, collision tumor, carcinosarcoma, and polypoid tumor, have been applied, ascertaining the divergent understanding of histogenesis of the spindle cell component. However, recently, the monoclonal theory has been accepted, wherein a single stem cell gives origin to both the cell components after further de-

differentiation or metaplastic alteration.^[8]

SpCC primarily occurs in males between the sixth and eighth decades of life, affecting the larynx most often.^[6, 7] In the case under discussion, SpCC was arising from the tongue, which is a comparatively rare location making it even rarer to present at a younger age. Possible risk factors include tobacco use, poor dental hygiene, alcohol abuse, and previous radiation therapy in the area.^[2, 4, 7]

In the case under discussion, the patient was a chronic alcoholic and tobacco chewer and had undergone ionizing radiation therapy 4 years back for carcinoma of the right

lateral border of the tongue.

Histologically, the mesenchymal component in SpCC predominates, forming the bulk of the tumor. The spindling morphology of the tumor cells is because of the lack of expression of cell adhesion molecules like e-cadherins and the resulting alteration in the keratin filament network.^[3, 4] I.H.C. study of our case suffices to the diagnosis, wherein the spindle cells were strongly positive for vimentin and focally positive for cytokeratin, while cytokeratin remained diffusely positive for squamous cells. These results may explain the morphological and functional properties of these malignant epithelial cells undergoing metaplastic changes, which results in loss of keratin.^[3]

The 2015 W.H.O. classification of oral cavity tumors has placed this tumor under the malignant epithelial tumors of S.C.C. and characterized it as SpCC.^[3] Disease progression in SpCC is characterized by relapses and metastases; wherein regional metastasis is more common than distant metastasis.^[2, 11] SpCC behaves more aggressively than S.C.C. per se, resulting in overall lower survival.^[3, 6] The prognosis of SpCC depends upon the tumor growth pattern, the depth of invasion, the presence or absence of vascular invasion, and regional or distant metastases, along with the history of radiation exposure.^[3, 6, 12]

Conclusion

Diagnosing SpCC is a histopathological challenge wherein I.H.C. studies remain the mainstay to overcome difficulties in the diagnosis. Although the tongue is an uncommon site for spindle cell carcinoma, however, during microscopy, a differential diagnosis of SpCC should always be kept in mind if malignant spindle cells are seen, and a thorough microscopic search for the epithelial component should be done. Ancillary techniques could aid in the final diagnosis by clearing the doubts, thereby improvising the multimodality treatment approach helping in the improved survival of patients with oral squamous cell carcinoma.

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None.

Conflict of interest

None.

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Ethics statement

Prior permission was taken from the Institutional Ethics Committee (DPU-IEC) to conduct this study.

References

1. Gupta R, Singh S, Hedau S, Nigam S, Das BC, Singh I, et al. Spindle cell carcinoma of head and neck: an immunohistochemical and molecular approach to its pathogenesis. *J Clin Pathol.* 2007;60(5):472-5.
2. Viswanathan S, Rahman K, Pallavi S, Sachin J, Patil A, Chaturvedi P, et al. Sarcomatoid (spindle cell) carcinoma of the head and neck mucosal region: a clinicopathologic review of 103 cases from a tertiary referral cancer centre. *Head Neck Pathol.* 2010;4(4):265-75.
3. Parikh N, Desai N. Spindle cell carcinoma of the oral cavity: a case report of a rare entity and review of literature. *J Adv Oral Res.* 2011;2(2):31-6.
4. Biradar MV, Dantkale SS, Abhange RS, Kamra HT, Birla K. Spindle cell carcinoma of the tongue: a rare variant of squamous cell carcinoma. *Ecancermedicalscience.* 2014;8:447.
5. Romãnach MJ, Azevedo RS, Carlos R, de Almeida OP, Pires FR. Clinicopathological and immunohistochemical features of oral spindle cell carcinoma. *J Oral Pathol Med.* 2010;39(4):335-41.
6. Su HH, Chu ST, Hou YY, Chang KP, Chen CJ. Spindle cell carcinoma of the oral cavity and oropharynx: factors affecting outcome. *J Chin Med Assoc.* 2006;69(10):478-83.
7. Wenig BM. Squamous cell carcinoma of the upper aerodigestive tract: precursors and problematic variants. *Mod Pathol.* 2002;15(3):229-54.
8. Choi HR, Sturgis EM, Rosenthal DI, Luna MA, Batsakis JG, El-Naggar AK. Sarcomatoid carcinoma of the head and neck: molecular evidence for evolution and progression from conventional squamous cell carcinomas. *Am J Surg Pathol.* 2003;27(9):1216-20.
9. Weidner N. Sarcomatoid carcinoma of the upper aerodigestive tract. *Semin Diagn Pathol.* 1987;4(2):157-68.
10. Batsakis JG, Suarez P. Sarcomatoid carcinomas of the upper aerodigestive tracts. *Adv Anat Pathol.* 2000;7(5):282-93.
11. Oktay M, Kokenek-Unal TD, Ocal B, Saylam G, Korkmaz MH, Alper M. Spindle cell carcinoma of the tongue: a rare tumor in an unusual location. *Patholog Res Int.* 2011;2011:572381.
12. Koseoglu RD, Sertçelik A, Ayva Y. A rare variant of squamous cell carcinoma of the tongue; spindle cell carcinoma. *J Ankara Univ Facul Med.* 2005;58:11-4.