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Recurrent spindle cell carcinoma of tongue- report of a rare case in an unusual location

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Abstract
Spindle cell carcinoma (SPCC) is a rare variant of squamous cell carcinoma having rapid growth and recurring nature. The scarcity of available literature, suggests that spindle cell carcinoma of the tongue is extremely rare. We present a 68-year-old male with a 1.5 x 1cm, recurrent, and rapidly proliferative lesion of the tongue for 2 months duration. We performed wide local excision with ipsilateral selective neck dissection. Histopathology and Immunohistochemistry confirmed sarcomatoid squamous cell carcinoma. He remains disease-free since 12 months of surgery. SPCC should be always considered during workup for any exophytic recurrent lesion from the tongue. Such lesions should be excised locally with a histologically negative margin and neck dissection for prevention of recurrence.

Keywords: spindle cell carcinoma, squamous cell carcinoma, lane tumor, carcinosarcoma

Introduction

The sarcomatoid variant of squamous cell carcinoma is an aggressive type of sarcoma comprising 3% of all squamous carcinomas of the head and neck region [1]. It is a peculiar, bimorphic malignant tumour made up of epithelial and mesenchymal components, accounting for around 1% of all oral cancers [2, 3].

To the best of our knowledge, less than 15 cases of spindle cell carcinoma of the anterior two-thirds of the tongue have been reported in world literature. Therefore we present this rare tumour with an unusual location and recurring tendency to contribute in part to the better understanding and awareness of this tumour.

Case presentation

A 68-year-old gentleman, a chronic alcoholic and a chronic smoker presented to our surgical oncology OPD with a recurrent, rapidly growing, painless exophytic growth over the left side of his tongue for the past 2 months.

Clinical history revealed that he had a nodular lesion in the left lateral border of the tongue which was excised locally and the biopsy was inconclusive. The lesion has recurred within 3 weeks and undergone re-excision which exhibited malignant spindle cell neoplasm and immuno-reactivity to Tumor protein 63 (P63), Smooth Muscle Actin (SMA), S100. Within 6 weeks following previous surgery, he had developed a recurrent exophytic growth at the same site. The lesion was not associated with bleeding or ear pain. There was no history of neck swelling or difficulty in swallowing or speech.

Physical examination revealed a 1.5 x 1 cm nodular, non-tender proliferative lesion arising from the left lateral border of anterior two-thirds of the tongue with adjacent leuokoplakic patch, it was located at 3 cm from sulcus terminalis and 5cm from tongue tip (Figure-1A). No extension of in-duration beyond the lesion or to the floor of the mouth, there was no tenderness, trismus, ankyloglossia or tongue deviation. There was no palpable cervical lymphadenopathy. The lesion was clinically staged as T1 N0 M0.

Biopsy was reviewed at our centre and confirmed as a cellular spindle cell lesion. Ultrasonography neck showed bilateral benign cervical nodes. As the patient was claustrophobic, Magnetic resonance imaging (MRI) head and neck could not be done. Computed Tomography (CT) chest ruled out lung metastasis. A multidisciplinary tumour board discussion was done and it was decided to proceed with surgery. He underwent wide local excision of the tumour with 3 dimensionally 1 cm margin (Figure-1B), Intraoperative histopathology frozen section confirmed as margin were free. Primary closure of tongue and left side extended supra- omohyoid neck node dissections were done. Histopathology showed cellular spindle cell lesion with severe dysplastic adjacent stratified squamous epithelium with infiltrating spindle cell (Figure-2). Resected margin and neck node were histologically free of neoplasm. Immunohistochemistry demonstrated positivity to Cytokeratin 5/6 (Figure-3), vimentin, SMA, and occasional cells positivity to P40 (Figure-4) and 40-50% of Ki67 (Figure-5). It was compatible with spindle cell (sarcomatoid) squamous cell carcinoma.

After a multidisciplinary tumour board discussion, it was decided to keep close observation and he remains disease-free since 15 months of surgery.

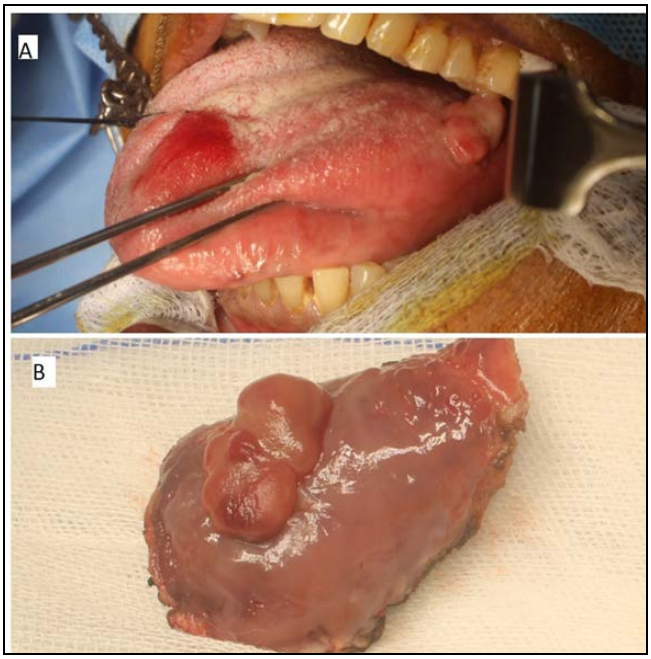

Fig 1: showing a 1.5 x 1 cm exophytic growth over the left lateral border of anterior 2/3rd of the tongue (A) and Gross resected specimen (B).

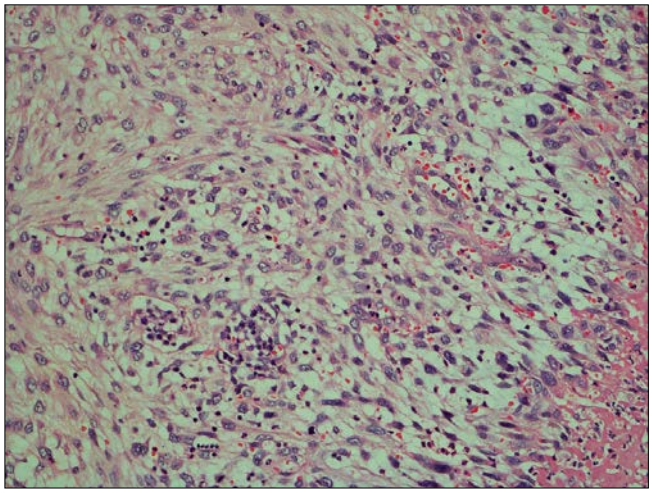

Fig 2: Microscopy of resected specimen (20x- Power) showing sheets of infiltrating pleomorphic spindle cell and atypical mitotic figures.

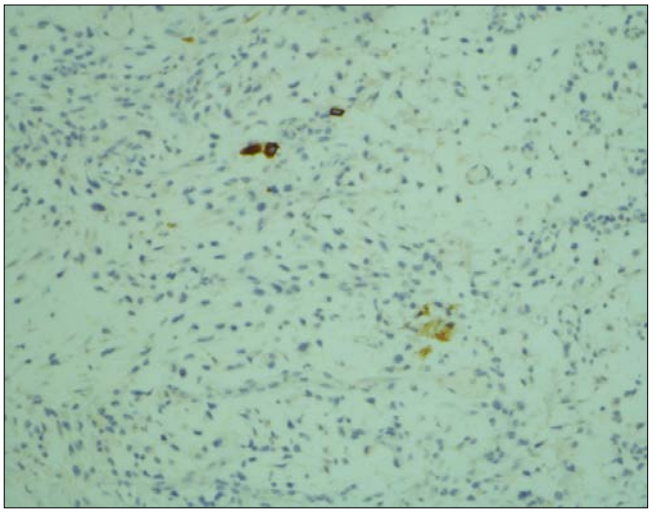

Fig 3: Immunohistochemistry demonstrated positivity to Cytokeratin 5/6.

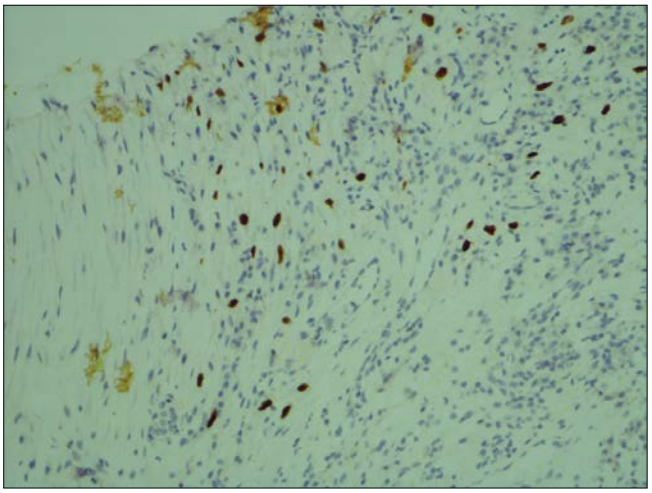

Fig 4: Immunohistochemistry demonstrated positivity to P40. (20x-Power)

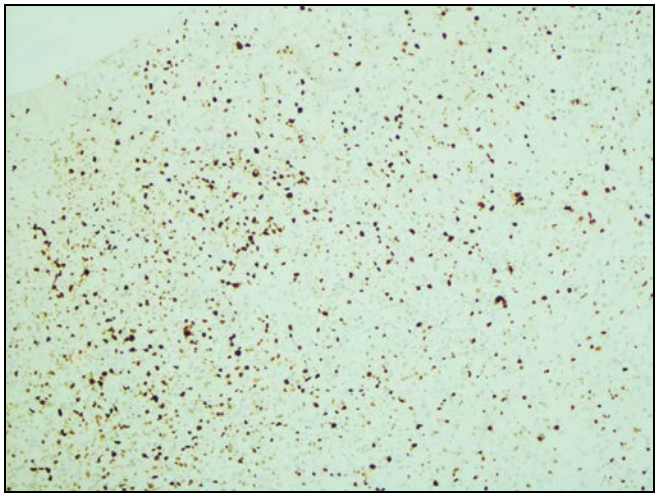

Fig 5: Immunohistochemistry demonstrated positivity to ki-67 40-50%. (10x-Power)

Discussion

SPCC was first explained by Virchow in1865[3], and characterized by predominantly invasive spindles cells in conjunction with dysplastic squamous epithelial islands which simulate a true sarcoma but are epithelial origin[4]. SPCC also called as, "sarcomatoid carcinoma", "carcinosarcoma", "pseudosarcoma", "pleomorphic carcinoma", "polypoid carcinoma", "collision tumor', "metaplastic carcinoma", and "Lane tumor", which itself reflects uncertain histogenesis and behavior of the tumor [3].

It classically presents as a pedunculated, polypoidal mass, occasionally nodular and fungating mass or ulcer affecting elderly people between 5th to 7th decade of life with male predominance [1]. It commonly originates from the upper aerodigestive tract like larynx, hypopharynx, oesophagus, scalp, oral mucosa [5]. In our case, the tumour was arising from the anterior lateral border of the tongue, which is a very rare location chronic smoking, alcoholism and poor oral hygiene are the potential risk factors [1]. This rare entity grows rapidly, recurs frequently and is prone to metastasis early to cervical lymph nodes and lung [1, 3, 5].

Diagnosis is often challenging. The presence of malignant undifferentiated spindle cell proliferation along with in situ or invasive squamous components is essential for histo-morphological diagnosis.

Demonstration of immuno-histochemical markers like cytokeratin and vimentin co-expression and Epithelial Membrane Antigen (EMA) on the spindle cell component helps to confirm the diagnosis by differentiating SPCC from other sarcomatous lesions [6]. Immunoreactivity for Ki-67, SMA and p63 is useful for some cases [6].

Staging workup includes CT chest and MRI neck often needed. Despite the poor prognosis of SPCC, Wide local excision with a safety margin and neck dissection with or without radiotherapy have a better survival rate than radiation alone [3, 5].

Overall survival mainly depends on the depth of local invasion, vascular invasion, tumour grade, lymph node status, distant metastasis or recurrence [7].

Our patient had classical fast-growing nodular growth on the anterior tongue with multiple recurrences. Histology showed cellular spindle cell lesion with dysplastic adjacent squamous epithelium and immunoreactivity for Cytokeratin 5/6, SMA and vimentin were eased in confirming spindle cell carcinoma of the tongue. With the wide local excision and Ipsilateral selective neck dissection, He remains disease-free at 12 months following surgery and he kept on close follow up.

Conclusion

Spindle cell carcinoma is a rare variety of squamous cell carcinoma and the tongue is the very unusual location of its origin. It tends to grow rapidly, and it has a high local recurrence. Therefore, SPCC should be always considered during workup for any exophytic recurrent lesion from the tongue. Such lesions should be excised locally with a histologically negative margin and neck dissection for prevention of recurrence.

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