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Lane Tumor with lame prognosis: An unusual case report.

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Abstract

According to WHO 2020, Lane tumor commonly called as Spindle cell squamous carcinoma (SCSC) is classified under malignant epithelial tumors, a monoclonal dedifferentiated form of conventional squamous cell carcinoma accounting only 1%. Though a rare variant, it represents an aggressive bimorphic/biphasic neoplasm exhibiting poor survival outcome, increased metastasis and recurrence. A 67-year-old male reported with an ulcerated nodular growth in the oral cavity, diagnosed as SCSC on incisional biopsy. It was T4N1M0. The management call was neo-adjuvant chemotherapy instead of surgical resection with radical neck dissection as it was too morbid. Unfortunately, the patient expired within 2 months of diagnosis before completing the planned chemotherapy cycles. This unusual case is documented because of sarcomatoid component which can mislead histopathological diagnosis with various other similar looking reactive, benign and malignant lesions and its worst prognosis, making SCSC one of the most challenging and interesting of all the head and neck tumors.

Keywords: Lane Tumor, Oral cavity, Neo-adjuvant chemotherapy.

Introduction

According to World Health Organization, 2020 Spindle cell squamous carcinoma (SCSC) is a variant of squamous cell carcinoma (SCC) characterized by predominant malignant spindle and/or pleomorphic cells.[1] Now, it is regarded as epithelial-mesenchymal transition undergone SCC.[2] Based on the microscopic peculiarities, SCSC is recognized with numerous alternate terminologies, like Sarcomatoid SCC, Carcinosarcoma, Pseudosarcoma, Polypoid SCC, Lane tumor, Collision tumor[3,4], Pleomorphic carcinoma and Metaplastic carcinoma.[4] Commonly seen in the larynx than in the oral cavity and oropharynx, accounts for < 1% affecting elderly males. SCSC linked to cigarette smoking, alcohol consumption and radiation exposure grows as a polypoid mass often with surface ulceration.[1] The article aims to report

this rare entity in the oral cavity with keen follow-up showing worst prognosis.

Patient Information

A 67-year-old male reported with a chief complaint of pain and intraoral growth over lower right back region of jaw since 2-3 months. The growth had increased to attain the present size, which developed after self-removal of tooth with slight force because of mobility. There was pain which was continuous in nature and moderate in intensity and radiated to the right ear and right cervical region. History of bleeding from the growth on brushing, difficulty in mastication and burning sensation was noticed. There was no history of trauma. Patient had habit of bidi smoking since 20-25 years, 6 times a day (1 packet = 6 Bidis). Habit of tobacco quid keeping in lower right lingual vestibule after dinner before going to bed and sleeping without spitting was also noted. Patient was diabetic since 2-3 years and was under medication (Glycomet). His past dental and family history were non-contributory.

Clinical Description

There was no facial asymmetry seen on extra-oral examination. Right submandibular lymphnode was freely movable and tender on palpation. Intra-oral examination revealed ulcerated exophytic nodular growth over the right posterior alveolus covering the occlusal aspect of 48 to retro-mandibular region and extending into the lingual and buccal vestibule. It was approx. 3.5 x 2 cm, irregular shaped, reddish pink covered with yellowish pseudomembranous slough. On hard tissue examination, teeth present were 11, 12, 34, 48. Severe gingival recession showing Grade II mobility was seen with 11, 12, 34, 48. [Figure 1]

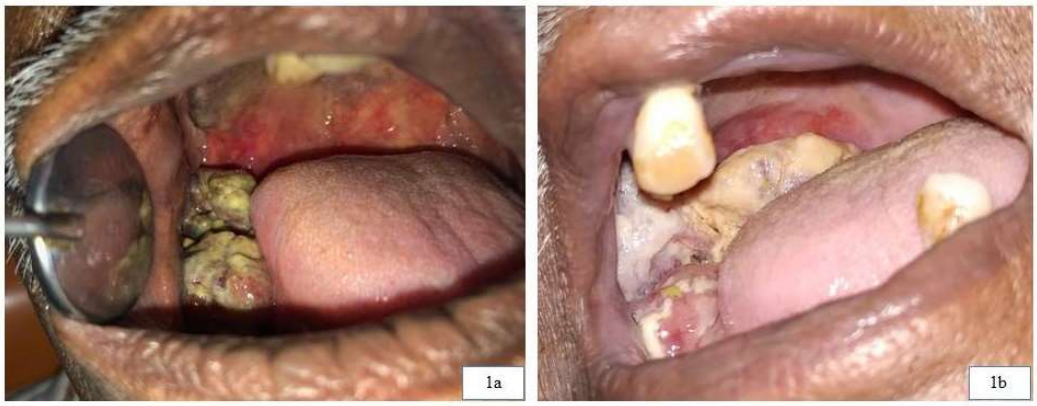

Figure 1 (Intra-oral examination) 1a: First visit. **1b:** Five days before patient expired.

Diagnostic Assessment / Investigations

In accordance with the history and clinical evaluation, the provisional diagnosis of malignancy involving the lower right alveolus was made. Radiographic examination on orthopantomogram, revealed peri-apical radiolucency with loss of lamina dura with 48, severe bone loss with 34, 48 and horizontal bone loss with 11, 12. Radiological diagnosis of generalized severe periodontitis was made.

The 18 F-Fluorodeoxyglucose Positron emission tomography (18 F-FDG PET CT) revealed metabolically active enhancing soft tissue thickening involving the right side of soft palate extending to the posterior part of right upper alveolus, right upper gingivobuccal sulcus, right retromolar trigone, anterior pillar of right tonsil, right lower gingivoglossal sulcus and right tonsillolingual sulcus, with focal erosion of the adjacent part of right mandible at right third molar tooth and infiltration into the sublingual soft tissue and the floor of the mouth- consistent with the known primary neoplastic pathology. Metabolically active right level II cervical lymph node appeared metastatic. [Figure 2]

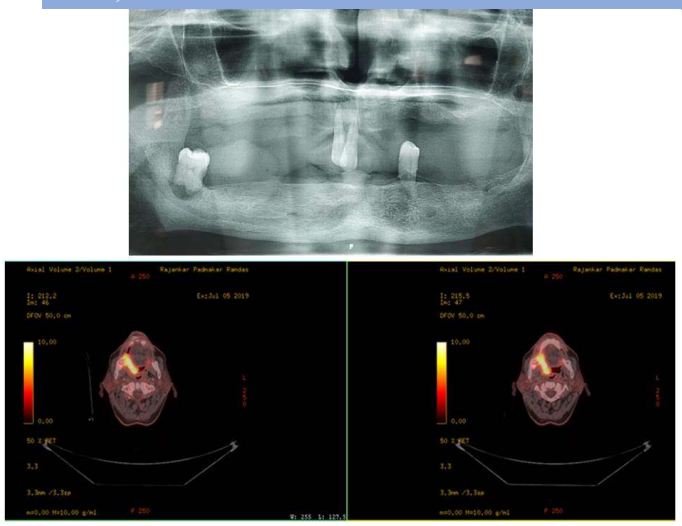

Figure 2: Radiographic examination – OPG & 18 F-FDG PET CT.

Blood investigations revealed Hemoglobin - 12.4 gm/dl, Total Leucocyte Count -14100/cumm, Red Blood Cell – 4.51 millions/cumm and Platelet count – 4.76 lakh/cumm. In Differential Leucocyte Count, Neutrophils were high - 81% and Lymphocytes were low - 13 %. Random Blood Sugar level was 194 mg/dl.

Incisional biopsy was done, macroscopic examination showed two bits of soft tissue specimens - brownish white in color, soft to firm in consistency and of size 1.1 x 0.8 cm and 0.9 x 0.8 cm. [Figure 3]

On histopathological examination, lesional tissue showed overlying thin parakeratinized ulcerated stratified squamous epithelium with epithelial dysplasia. Underlying connective tissue composed predominantly of fascicles of anaplastic spindle shaped cells. Few dysplastic epithelial islands with keratin pearl formation were seen. The cells in fasciculated pattern were elongated and showed features like marked cellular & nuclear pleomorphism, hyperchromatism, altered nuclear cytoplasmic ratio and atypical mitotic figures [Figure 4a–10x, 4b–20x, 4c–40x]. In accordance with the clinical, radiological and histopathological evaluation, the final diagnosis of Spindle cell squamous carcinoma was made. For further confirmation the specimen was sent for Immunohistochemistry (IHC) – Cytokeratin, p63, EMA, CEA, SMA, Vimentin, Desmin but the results were not specified.

Figure 3: Macroscopic examination.

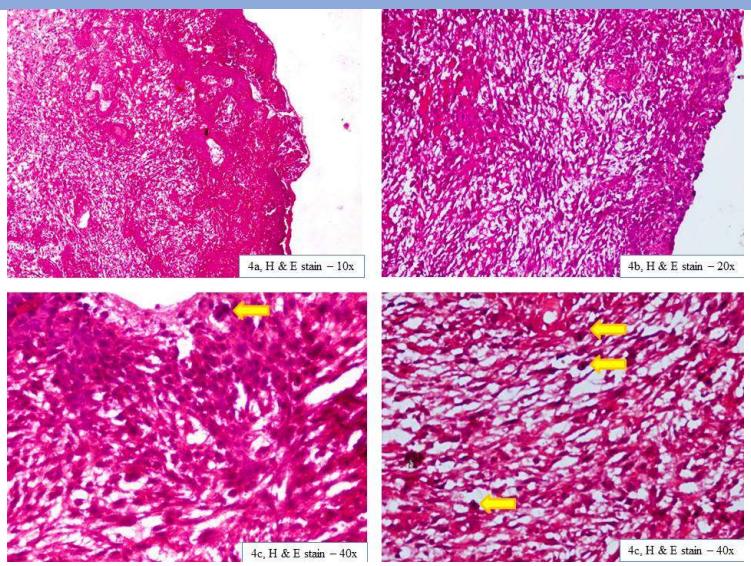

Figure 4a: Overlying thin parakeratinized ulcerated stratified squamous dysplastic epithelium, H & E stain – 10x. **4b:** Lesional connective tissue showing fascicles of anaplastic spindle shaped cells, mitotic figures and keratin pearl formation, H & E stain – 20x. **4c:** Dysplastic features like marked cellular & nuclear pleomorphism, hyperchromatism, altered nuclear cytoplasmic ratio and atypical mitotic figures (arrow), H & E stain – 40x.

Therapeutic Intervention

Considering the extensive involvement of the lesion, surgical resection with radical neck dissection was too morbid, therefore the management call was for neo-adjuvant chemotherapy. Patient was then referred to Regional Cancer Hospital & Research Centre, where total 6 chemotherapy cycles were planned. Chemotherapy drugs prescribed were Inj. Pacilitaxel [100mg] with 1 point NS over 3 hrs (45 drops/min), Inj. Cisplatin [50mg] with 1 point NS over 2 hrs (65 drops/min), Inj. 5-Flurouracil [500mg] with 1 point NS over 2 hrs (65 drops/min).

Follow-up and Outcomes

Knowing the aggressive nature of the tumor and its worst prognosis, the patient was kept under follow-up every month. Patient underwent 5/6 chemotherapy cycles. Intra-oral examination showed the extension of lesion reaching the midline of the soft palate [Figure 1]. Patient expired within a span of 2 months of diagnosis.

Review of Literature

Table 1: Review of literature of Spindle cell squamous carcinoma in orofacial region over a decade.

Authors	Date of Publication	Age/ Gender	Clinical presentation / Site	Symptoms	Radiographic finding	IHC markers	Treatment
Samuel S et al[5]	2013	52/F	Maxillary posterior region	Mobility of upper anterior teeth	Loss of bony architecture with floating teeth appearance	-Creatine kinase ++ - Vimentin ++ - S-100 + - EMA – - HMB45 – - SMA –	Loss of follow-up
Ezulia T et al[6]	2015	33/F	Large fungating mass over right Mandibular posterior region	Pain, odynophagia and trismus with occasional episodes of bleeding	CT showed a lytic lesion	- EMA + - Vimentin + - CK AE1/AE3 – - CK 7 –	Segmental mandibulectomy
Chaudhary M et al[7]	2015	60/M	Ulcerative lesion in Mandibular anterior region	Dull aching pain with burning sensation	Irregular radiolucency with floating tooth appearance	- Vimentin ++ - CK +	Loss of follow-up
Patankar S et al[8]	2016	38/M	Soft, erythematous gingival swelling in Mandibular anterior region	Painful swelling	Vertical bone loss	- AE1/AE3 ++ - EMA + - S-100 – - Myogenin – - Desmin – - p63 –	Loss of follow-up
Xie L et al[9]	2017	74/F	Mandibular posterior region	Painful swelling	Low-density shadow	-Creatine kinase + - Vimentin + - S-100 – - EMA – - Desmin – - CD31 – - CD34 – - Bcl-2 – - GFAP –	Surgical resection and titanium plate prosthesis

Mahajan A et al[10]	2017	51/M	Soft tissue lobular growth in the mandibular posterior region	Mobility of mandibular left third molar	Well-defined arc-shaped osteolytic lesion with noncorticated Borders	- EMA + - Vimentin + - S-100 – - Desmin – - CD45 – - CD31 – - CD34 –	Surgery followed by chemotherapy
Reddy S et al[11]	2017	48/M	Polypoid lesion of maxilla	Not specified	CBCT showed opacification and calcifications	- Vimentin + - Pancytokeratin –	Not specified
Wadhawan R et al[12]	2018	50/F	Mandibular anterior region	Sickle shaped painful swelling	Loss of trabecular pattern	- AE1/AE3 ++ - EMA + - S-100 – - Myogenin – - Desmin – - p63 –	Loss of follow-up
Panda S et al[13]	2018	42/M	Ulceroproliferative lesion in left side of lower gingivobuccal sulcus	Painful swelling	Buccal cortical bone loss	- Vimentin + - SMA –	Not specified
Rai H et al[14]	2018	45/F	Left buccal mucosa	Painful swelling	Not specified	-Pancytokeratin +	Surgical excision
Varshini M et al[15]	2019	62/M	Maxilla	Swelling and discharge		-Pancytokeratin +	Radiotherapy
Colney L et al[16]	2022	62/M	Polypoidal growth at tip of the Tongue	Painless mass	Not specified	- CK + - p40 + - EMA – - CD34 – - BCL2 – - SMA –	Surgical excision with adjuvant radiation therapy
Mohanty A et al[17]	2022	48/M	Lateral border of the tongue	No pain or discharge	Not specified	- CK+ - Vimentin +	Resection
Sobiech L et al[18]	2023	45/M	Maxillary anterior region	Unstable tooth	Extensive blurred area of osteolysis	Not specified	Surgical excision with reconstru

							ction and combined chemotherapy and radiotherapy

Epithelial Membrane Antigen (EMA), Human melanoma black 45 (HMB45), Cytokeratin (CK), Smooth Muscle Actin (SMA), Glial fibrillary acidic protein (GFAP).

Discussion

Lane tumor commonly known as Spindle cell squamous carcinoma is composed of pleomorphic or malignant spindle cells with SCC component. It is derived from the squamous epithelium with the ability to produce mesenchymal intermediate filaments and demonstrates divergent differentiation by epithelial-mesenchymal transition.[1,3] This is because it contains remnants of dysplastic squamous epithelium and shows areas of transition to pleomorphic or malignant spindled cells. Some investigators have hypothesized that a dysfunctional change occurs at cadherin-caterin complex which is essential for intercellular adhesion results in shift or transition of tumor cells from squamous to spindle type with increased infiltration causing aggressive behaviour. Some cases of SCSC may develop after radiotherapy for a more differentiated SCC, a phenomenon called as "Dedifferentiation".[3,17] SCSC occurs most frequently in the larynx, especially the glottis with airway obstruction or hoarseness.[2] Cytological picture of metastatic SCSC often shows focal keratinizing SCC but in some cases a malignant spindle cell component is also observed. The diagnosis is straightforward which rests on demonstrating epithelial differentiation i.e. foci of conventional SCC mixed with sarcomatoid component or squamous dysplasia of residual surface epithelium. In addition to histopathological studies, immunohistochemical analysis can be carried out for cytokeratins AE1/AE3, EMA, p63, p40. However, 1/3rd of SCSC are purely spindled, a significant subset is negative for epithelial markers. Mesenchymal markers like vimentin, desmin, S-100, Osteopontin, bone morphogenetic protein etc. are considered [Table 1]. Genetic profile of SCSC harbours complex genetic alterations which is similar to poorly differentiated SCC.[1,2]

Differential diagnostic possibility can be a number of other malignant tumors like Squamous cell carcinoma, Intraosseus carcinoma, Chondrosarcoma, Myofibroblastic sarcoma, Fibrosarcoma, Malignant fibrous histiocytoma, Leiomyosarcoma, Rhabdomyosarcoma, Malignant peripheral nerve sheath tumour, Osteosarcoma, Mesenchymal chondrosarcoma, Kaposi's sarcoma, Angiosarcoma, Synovial sarcoma, , Fibromatosis, Leiomyoma, Nodular fasciitis, Reactive epithelial proliferations and Malignant melanoma.[5,7]

SCSC grows rapidly, typically diagnosed at a late stage and tends to metastasize early.[15] The treatment of the choice is surgical resection with radical neck dissection when clinically positive nodes are present. Adjuvant radiotherapy may be of benefit if surgical margins are positive. The role of chemotherapy is not published, but it may decrease the incidence of metastasis and recurrence of primarily sarcomatous lesion.[18] The 5 year disease-free survival rate for oral lesions is approximately 30%. Most deaths occur within 1 year of diagnosis.[3] The prognosis of oral SCSC is worse than SCSC arising in other anatomic sites such as larynx, hypopharynx etc. but it is similar to the prognosis of conventional SCC.

Timeline of History

Briefly outlined in Flowchart below.

February 2019	Patient experienced pain & growth in right posterior mandible.
May 31st, 2019	Patient reported in OPD.
June 12th, 2019	Histopathological diagnosis reported Spindle cell squamous carcinoma.
July 5th, 2019	18 F-FDG PET CT scan.
July 8th, 2019	Management call for neo-adjuvant chemotherapy.
July 12th, 2019	1st chemotherapy cycle.
August 28th, 2019	Patient expired.

Conclusion

Though SCSC occurring in oral cavity is a rare entity, but many cases are been reported over a past decade. The aggressive nature of this Lane tumor leads to very poor/lame prognosis. IHC remains helpful for the confirmatory diagnosis. Early diagnosis and prompt intervention is must for aiding in better patient management and prognosis.

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