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Research Article

Mucosal ascc of oral cavity: a case report implicating need for aggressive therapy and close follow up

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A B S T R A C T

Acantholyticsquamous cell carcinoma is a rare variant of SCC and accounts for 2-4% of all SCC. Although, this lesion is commonly found on sun-exposed skin of head and neck, it is rarely seen in oral cavity. The acantholytic activity in tumor cells, high incidence of neck nodal metastasis and high loco-regional recurrence rates makes it different from other SCC variants in terms of long-term survival. We report one such case of oral ASCC in a 61-year-old male with recurrence after first therapy and being followed up till date after second surgery. So far this report documents the longest follow up of the patient with ASCC after therapy and emphasize the need for close follow-up, in addition to aggressive therapy for long-term disease-free survival.

INTRODUCTION

The oral squamous cell carcinoma (OSCC) is one among globally spread cancers and is the most common malignancy of head and neck region. The majority of squamous cell carcinoma cases are of conventional type as compared to its other morphological variants such as spindle squamous cell carcinoma, basaloid squamous cell carcinoma, adenosquamous cell carcinoma and papillary squamous cell carcinoma.[1]

Acantholytic (adenoid) Squamous Cell Carcinoma (ASCC) which is also known as pseudovascular SCC (gland-like features),angiosarcoma-likeSCC, and pseudovascular adenoid SCC is a rare variant of oral SCC. They are characterized by tumor cell acantholysis, pseudoluminand glandular differentiation patterns.[3]ASCC was first described by Lever (1947) as a malignancy affecting sweat glands. The recognized risk factors include ultraviolet radiation exposure and tobacco abuse.[4] Oral ASCC is reported to be a malignancy with poor prognosis and is often challenging to achieve long-term disease-free survival.[4,5]

This paper highlights a rare presentation of ASCC as an ulcero-proliferative growth on palate, in a patient with no exposure to known risk factors of oral SCC.

Also, this paper emphasises on the need for adjuvant therapy. irrespective of tumour size and the importance of aggressive post-therapy follow-up with review of literature.

Case Report

A 61-year-old male was referred to our outpatient department on December 29th 2010, with chief complaint of painless growth in the right palate for past one month. No associated personal habits were reported by the patient. Apart from the lesion, general physical condition of the patient was normal. Patient had history of angioplasty in 2005 and was on oral aspirin therapy, 75 mg once a day since then.

No abnormality was detected on extra oral examination. Intraoral examination revealed a brownish, well-defined ulcero-proliferative lesion in the right palate in relation to first premolar to first molar region measuring about 3 x 2.5cm, not crossing the midline. (Fig. 1) The growth was soft in consistency, non-tender and with increased tendency to bleed. Clinically, palpable right level IB node was evident on neck examination that was enlarged, painless, mobile and firm in consistency. In the absence of bony erosion on contrast enhanced CT images with 5mm cuts and no evidence of distant metastasis on imaging, the TNM staging was made as T2N1M0. Incisional biopsy was suggestive of well differentiated squamous cell carcinoma.

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Table 1 Cases of intraoral ASCC reported in the literature.

Reference	No. of cases	Mean Age	Gender	Location	Size (in cm)	L N metastases	Lesion	Therapy	Follow up (Months)
Kerawala *et al.* 2009	1	56	M	Lateral side of Tongue	1.6x1.1	Absent	Ulcer	TR,ND, RT	ROC (5) DOD (9)
Papadopoulou *et al.* 2010	1	72	F	Mandibular alveolar ridge	1.2 x0.5	NS	Irregular mass, with a central ulceration	TR,RT	ROC (10) DOD (17)
Yeoh MS *et al.* 2012	1	38	F	Buccal mucosa	6x5.4x2.8	Present	Ulcero-proliferative growth	TR, ND, CT, RT	ROC (5) DOD (7)
Gu *et al*.2012	3	70	M	Palatal mucosa	NS	NS	Ulcerative lesion	NS	ROC (48)
		61	M	Buccal mucosa	NS	NS	Ulcerative lesion	NS	NA
		38	F	Buccal mucosa	NS	NS	Ulcerative lesion	NS	DOD(7)
Shigeo Ishikawa *et al*.2014	1	64	F	Maxillary gingiva	6 cm in diameter	absent	Exophytic tumor	TR, B/L ND, Maxillectomy,Recon. With free radial forearmflapCT,RT	NED (30)
Xiaodong Hang *et al*.2020	1	54	F	Tongue	6x4x1	b/IIb LN	Exophytic lesion	TR, ND	NED (15)

NS – Not stated, NED – No Evidence of Disease, ROC – Recurrence of Cancer, RT -Radiation Therapy
NT – No Treatment, TR – Tumor Resection, DOD – Died of Disease, DOC – Died of Other Causes
ART – Adjuvant Radiotherapy, CT – Chemo Therapy, M – Male, F – Female.

The patient underwent right infrastructure maxillectomy and right extended supra-omohyiod neck dissection (level I to level IV). Split skin grafts were used to line the raw areas and patient was later rehabilitated with obturator. The surgical margins were clear with closest being medial margin that was 0.9cm away from the tumor. The final histopathology was suggestive of ASCC with 1 out of 19 nodes positive for metastasis and was at level IB. In view of pT2N1 status, the patient received ipsilateral radical radiotherapy of 60 Gy. The patient was asked to follow up regularly for once in every month in view of aggressive nature of the disease.

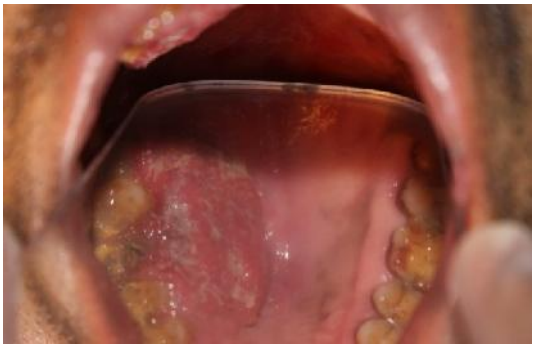

Fig 1 Intraoral photograph showing ulceroproliferative growth on the right palatal mucosa.

After 1 year and 8 months, on routine follow-up he presented with asymptomatic ulcero-proliferative growth in right lower gingiva of which he was unaware. On examination, there was an ulcero-proliferative growth extending from 44 to 46 again with high tendency to bleed. The contra lateral level 3 nodes were palpable. In view of suspected recurrence, PET CT was advised that was suggestive of metastatic active lesion in the right mandibular gingiva-buccal sulcus with SUV of 9.8 and lymphadenopathy at left level 2/level 4 with SUV of 4.2 and no evidence of distant metastasis. (Fig. 2)

Composite resection with contra lateral MRND and bi-paddle pectoralis major myo-cutaneous flap reconstruction was done. Histopathologic examination revealed tumor measuring 3.1x2x0.9 cm with free surgical margins. Microscopically, there were tumor islands with lobular pattern of keratinizing SCC and central regions containing rounded spaces(pseudo-glandular/alveolar areas) lined by polygonal cells with central lumen containing detached, dyskeratotic, acantholytic, neoplastic keratinocytes.

(Fig. 3) The patient refused to take radiotherapy for the second time even though he was in pT2N2C stage and preferred to be on regular follow up. He has been on regular follow-up for over 8 years and there are no complications or any signs of recurrence till date. (Fig. 4A, 4B, 4C)

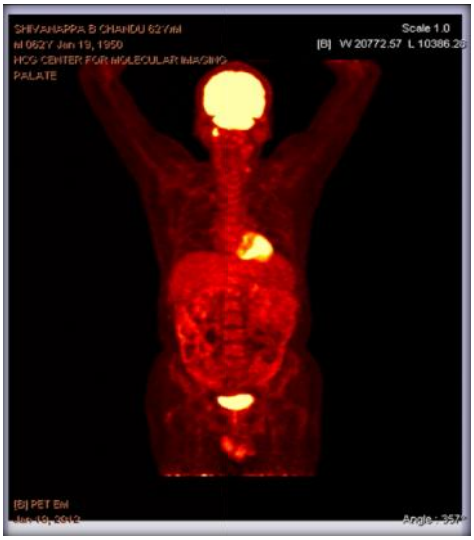

Fig 2 PET CT showing recurrent active lesion in the right mandibular gingiva-buccal sulcus with SUV of 9.8 and lymphadenopathy at left level 2/level 4 with SUV of 4.2.

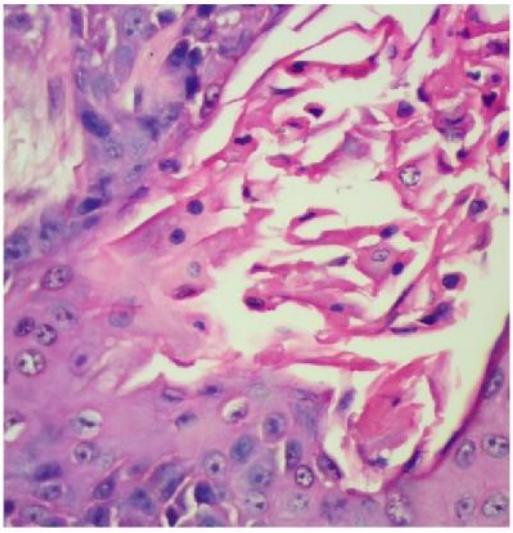

Fig 3 Photomicrograph showing island of keratinizing SCC with central region (pseudo-glandular/alveolar area) containing detached, dyskeratotic, acantholytic, neoplastic keratinocytes.

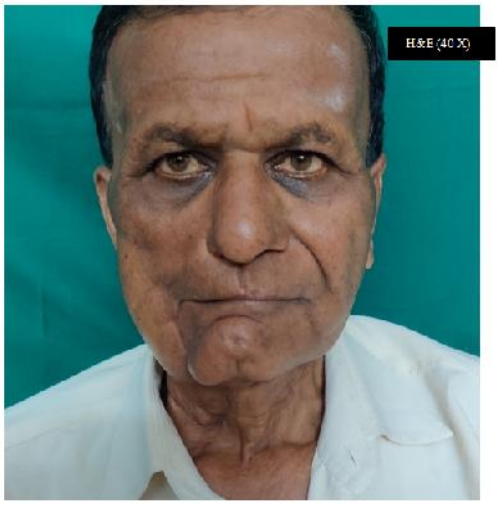

Fig.4A Front view on 10.12.2020. (8 years follow-up)

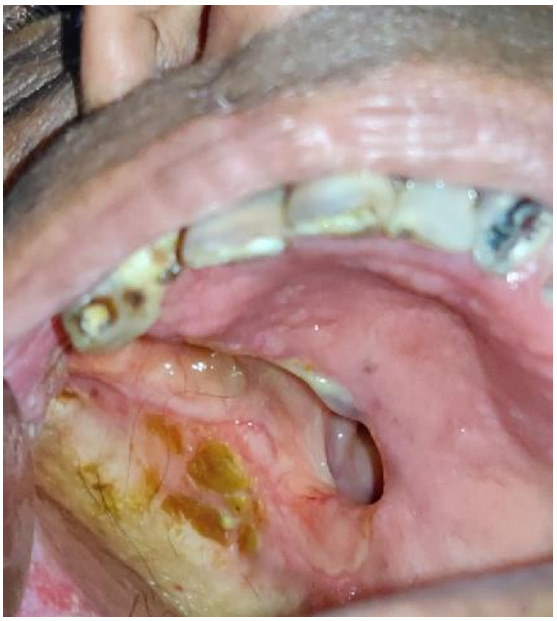

Fig. 4B Intraoral photograph of resection site (right palate) after first surgery. (8 years follow-up)

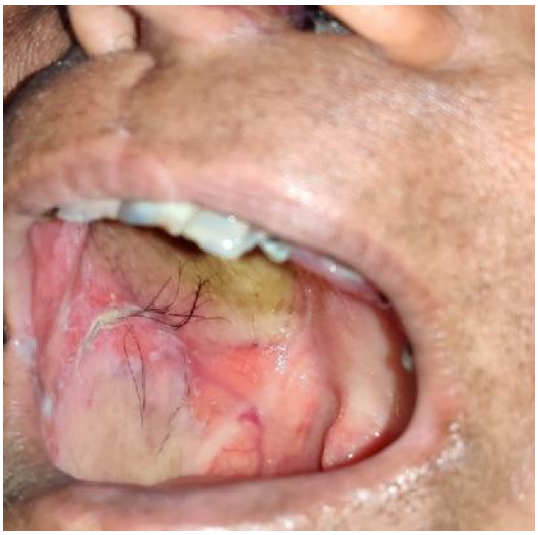

Fig 4C Intraoral photograph showing healing status of right buccal mucosa after second surgery.(8 years follow-up)

DISCUSSION

In 1947, Lever reported a neoplasm of sweat glands and termed it as adenoacanthoma which was later found to be of non-eccrine variant of squamous cell carcinoma(SCC). Acantholytic squamous cellcarcinoma (ASCC) isan unusual histopathological form of SCC, distinguished by its pronounced acantholysis and degeneration of cancer cells giving them pseudoluminalor pseudoglandular appearance.[1,5]It is commonly seen on sun exposed areas of the skin (head and neck of elderly men) and is found to have high recurrence and metastasis compared to SCC.

Histologically, ASCCis composed of conventional SCC cells with acantholytic degeneration in the tumor nests which gives it a pseudoglandular or pseudoluminal appearance. The glandular or ductular structures are bounded by epithelial cells alone or two layered cuboidal or polygonal cells comprising of a lumen with acantholytic and dyskeratotic cells. ASCC also contains different types of collagen fibers, spindle cell and inflammatory infiltrate in its connective tissue stroma.[2,3,6]

Histological criteria for diagnosing ASCC include a conventional squamous cell component (usually well-differentiated) with pseudolumina containing single or grouped acantholytic and dyskeratotic epithelial cells or cellular debris under the squamous neoplastic component.[3]This non-solid component usually shows an alveolar pattern or a pseudoglandular arrangement. All these characteristics were demonstrated by both lesions in the current case.

The differential diagnosis includes adeno squamous carcinoma, adenoid cystic carcinomas, mucoepidermoid carcinomas, angiosarcomas and conventional SCC with ductal involvement.[6]ASCC can be differentiated from adenosquamous carcinomas by lack of glandular structures and mucin stains negativity.[3] The absence of mucous cells and myoepithelial cells aids in rejecting adenoid cystic carcinoma and mucoepidermoid carcinoma. Also, ASCC resembles adenoid cystic carcinomas based on the appearance of glandular spaces and pretendence of fibrin as mucin, wherein actual ASCC elicits the glandular pattern which has an angular emergence. ASCC closely resembles angiosarcoma due to its similar histologiccharacteristics i.e., development of channels and intra- tumoral spaces.However, they differ in their clinical characteristics. Angiosarcoma comprises of hyperchromatic nuclei in endothelial cells.[3] On the contrary, ASCC have islands of dermal cells with projecting suprabasilar acantholysis. Angiosarcoma can be diagnosed by the presence of factor VIII, CD31 and CD34 antigens. Conventional squamous cell carcinoma differs from ASCC in terms of histological characteristics as well as its less aggressive nature.

In view of high tendency for local regional recurrence, we advised matrix metalloproteinase-8 (MMP-8) and matrix metalloproteinase-10 (MMP-10) assessment in the patient, which turned positive for both the markers. confirming the same.

To our knowledge, only 61 cases of ASCC have been reported in oral cavity till date and that includes the present case as well. Among those, lip is the most commonly affected site (41%) followed by buccal mucosa(13%) and alveolar ridge (13%). It has also been found on tongue, floor of the mouth, gingiva, alveolar ridge and buccal mucosa.[8]This malignancy has shown higher predilection to occur in males.[9]

For ASCC of skin and lip, the preferred choice of treatment is surgical excision. This treatment modality showed very low recurrence and metastasis.[10]In contrast to conventional oral SCC, ASCC in our patient was found in a patient without exposure to known risk factors, was associated with node positive neck on initial presentation and showed high recurrence rate in spite of clear surgical margins and aggressive adjuvant therapy. Also, the high regional metastatic potential was confirmed by positive MMP enzymes.

Although, the lesion was smaller in size, its propensity of metastasis to regional lymph nodes was high and hence there was a need for aggressive therapy. Also, present case which was found in oral cavity demanded a multi-disciplinary treatment that includes neck dissection, primary resection and reconstruction with PMMC flap followed by radiation therapy.

The prognosis of ASCC is different at various anatomic sites and the mucosal surfaces of the upper aero digestive tract and oral cavity has the worst prognosis.

CONCLUSION

The histological variants of oral SCC and their biological behaviour can also contribute to the prognosis of oral SCC in terms of recurrence or disease-free survival. Although, the current therapeutic guidelines for oral SCC are based on TNM staging, the therapeutic decision making should also consider histologic variants if they are known to the surgeon before surgery. Also, if the histologic variants are known on final histological examination of the resected specimen, the emphasis should be made on aggressive follow up of the patients in order to detect recurrences early.

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