

Non Healing Ulcer of Tongue: A Rare Entity

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Abstract

Pseudoglandular carcinoma or Acantholytic squamous cell carcinoma is an uncommon, histopathologically distinct variant of squamous cell carcinoma. ASCC commonly occurs in areas of skin exposed to sunlight and has only rarely been seen on mucosal surfaces such as the oral cavity. Although the World Health Organization has defined ASCC as an original entity, the imaging findings of ASCC have not been adequately described. We herein report a case of ASCC occurring in the oral mucosa with extensive review of past published cases.

Keywords: Ulcer, Tongue, Case Report, Tongue

Case report

A 50 year old female presented with non healing painful ulcer on the left side of tongue which she observed four months back. Patient further complained of burning sensation on affected area and left cheek region since one and half months. She also gave history of tobacco placement on left side in the mouth since one and half years and has quit the habit since three months. Patient revealed no relevant past medical or dental history and no family history. General physical examination revealed a moderately built and nourished, well oriented and cooperative patient. No signs of anemia or cyanosis were

observed. On extraoral examination no extraoral or TMJ abnormality was noted. Lymph nodes were found to be non palpable. On intraoral examination, a solitary tender ulcer with irregular margins, inverted edges, reddish to pink in colour on the left lateral margins of the tongue approximately 3x2 cm in size was found. The lesion extended from left permanent mandibular first molar to third molar, 0.5 cm from the midline of the tongue, covered by a whitish yellow membrane resembling necrotic tissue, and surrounding area shows distinct whitish discoloration. Palpation of the lesion revealed tenderness and marked induration. Palpation of buccal mucosa revealed loss of elasticity on the left side and fibrotic bands could be felt. Other findings included mild generalized stains and calculus, caries wrt 46 and grossly decayed 47. Clinical provisional diagnosis of oral squamous cell carcinoma was given. No relevant findings were observed in OPG. MRI examination revealed a focal ulceration along the left border of the tongue with hyper intense signal on T2/T2-SPiR images, rest of the tongue and midline raphe appeared normal; and a clinical histopathological correlation was advised in view of a non healing ulcer. Biopsy of the lesion was performed to proceed further with the diagnosis. Gross specimen received was a single soft tissue 1x1.2x0.6cm in size,

irregular in shape and creamish white in color. Histopathological examination revealed presence of sheets, cords and clusters of malignant epithelial cells within the connective tissue stroma. On higher magnification cellular malignant features like cellular pleomorphism, bizarre cells, nuclear hyperchromatism and multiple nucleoli with evident mitotic figures were observed. These malignant cells appeared in clusters with acantholysis and necrosis of central cells giving a distinct pseudo glandular appearance. Acantholytic change appeared to be a characteristic and diagnostic feature. Further, Keratin pearls and individual cell keratinization was also observed. The tumor stroma showed thin interlacing collagen fibers, endothelial lined blood vessels with mild to moderate chronic inflammatory infiltrate. After clinical, radiological and histopathological investigations, the final diagnosis of a rare variant-acantholytic squamous cell carcinoma was made.

Discussion

Acantholytic squamous cell carcinoma (ASCC) is an infrequent histopathologic variant of squamous cell carcinoma, characterized by acantholysis of the tumor cells, creating pseudolumina and false appearance of glandular differentiation. An uncommon but well-recognized variant, first described by Lever in 1947.^[1] ASCC has been reported to originate in the sun-exposed skin of the head and neck and in other sites, most commonly occurs on the lips. It is rare on mucosal surfaces of the upper aerodigestive tract, where there is

some suggestion that it might behave more aggressively than conventional squamous cell carcinoma, and it is associated with a poor prognosis.^[2] According to the International Classification of Diseases for Oncology, the ICD-O code for the following in 8075/3. This variant is also popularly referred to as adenoid squamous cell carcinoma (SCC), pseudo glandular SCC, SCC with gland-like features, Angiosarcoma-like SCC, Pseudo vascular adenoid squamous cell carcinoma.^[3] The term acantholysis, coined by Auspitz in 1881, is derived from the Greek words *akantha*, meaning a thorn or prickle, and *lysis*, i.e. loosening. Acantholysis is defined as the loss of coherence between epidermal cells due to the breakdown of their intercellular bridges. The cells remain intact but are no longer attached to each other; they tend to acquire the smallest possible surface area and become rounded up, resulting in intra-epidermal clefts, pseudoglands, vesicles and bullae. ASCC expresses cytokeratins but not glandular and endothelial cell markers, which distinguishes it from eccrine and endothelial neoplasms. Adenosquamous carcinoma signify a challenge in diagnostic routine for its rarity, varied array of clinical presentations and histological features, since the adenocarcinomatous component may be, at times, difficult to identify. The recognition of the specific subtype of squamous cell carcinoma is of great significance in prognosis and outcome. A thorough review of past published cases with its clinical parameters are tabulated below -

S. no.	Authors	Year	Patient age	Patient gender	Location of the lesion
1	Jacoway et al ⁴	1971	41-75	13 M/2F	upper and lower lip
2	Tomich and hutton ⁵	1972	50,53	2M	Lower lip
3	Weitzner ⁶	1974	67	M	Lower lip
4	Goldman ⁷	1977	61	M	Tongue
5	Takagi et al ⁸	1977	50,56	F,M	Gingiva, tongue

6	Caya et al ⁹	1985	50	M	lip
7	Jones et al ¹⁰	1993	42,47,58	F,M,M	Floor of the mouth, lower lip
8	Blackburn et al ¹¹	1999	78	F	Upper lip
9	Zidar et al ¹	2006	59,77	F	Buccal mucosa, floor of the mouth
10	Kasafuka et al ¹²	2006	64	F	Floor of mouth
11	Driemel et al ¹³	2008	50,68,57	M	Tongue Floor of mouth
12	Kerawala et al ²	2009	56	M	Lateral side of Tongue
13	Papadopoulou et al ¹⁴	2010	72	F	Maxillary alveolar ridge
14	Prasad et al ¹⁵	2010	70	F	GINGIVA
15	Yeoh MS et al ¹⁶	2012	38	F	Buccal mucosa
16	Terado et al ¹⁷	2012	73	F	Mandibular alveolar ridge
17	Nayak et al ¹⁸	2012	45,53	M	Floor of the mouth, maxillary ridge
18	Vidyavathi K et al ¹⁹	2012	40	M	Floor of the mouth
19	Patil SK et al ²⁰	2014	49	F	Buccal mucosa
20	Deepak et al ²¹	2014	38	M	Tongue
21	Kavita Mardi et al ²²	2014	50	M	-
22	Donthi D et al ²³	2014	38	M	Tongue
23	Thanakappan P et al ²⁴	2015	55	M	Buccal mucosa
25	Allon I et al ²⁵	2016	-	M	Lip
26	Lima CF et al ²⁶	2016	45	M	Hard palate
27	Lin JS et al ²⁷	2016	55	M	Soft palate
28	Custodio M et al ²⁸	2018	100	F	Hard and soft palate
29	Neto HS et al ²⁹	2018	71	M	Hard and soft palate
30	Chandrakala J et al ³	2018	63	F	Mandibular alveolar ridge
31	PRESENT CASE	2019	50	F	Tongue

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Legends Figure

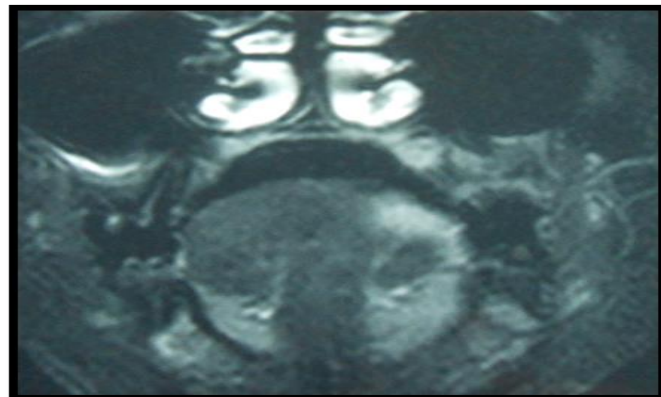


Figure 1: MRI image revealed a focal ulceration along the left border of the tongue with hyper intense signal on T2/T2-SPIR images

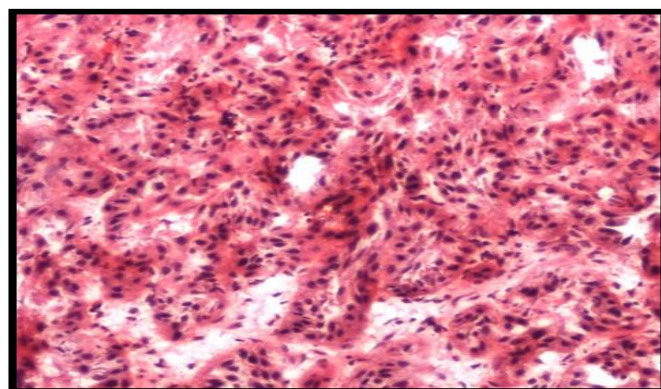


Figure 2: histopathological examination revealed the presence of sheets, cords and clusters of malignant epithelial cells within the connective tissue stroma

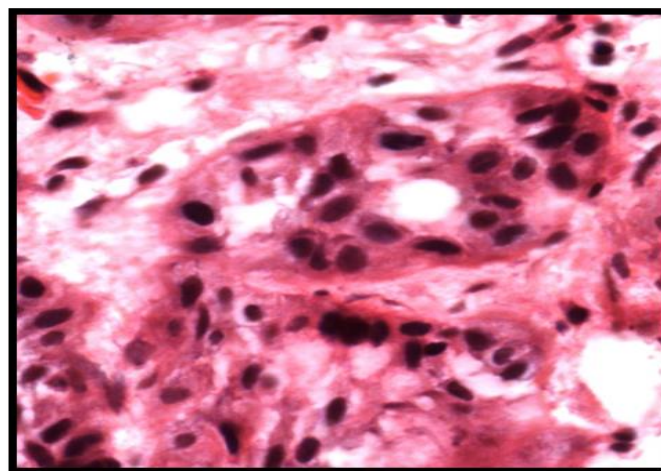


Figure 3: malignant cells appeared in clusters with acantholysis and necrosis of central cells giving a distinct pseudo glandular appearance.