Case Report

Adenoid Squamous Cell Carcinoma of Oral Cavity: a Case Report

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ABSTRACT

Adenoid squamous cell carcinoma is a rare variant of squamous cell carcinoma with features of adenoid pattern. It has been reported to originate in the sun-exposed skin of the head and neck region. Although rare, there are cases documented within the oral cavity and nasopharynx. The clinical behaviour and the prognosis are variable. We report a case of adenoid squamous cell carcinoma in a 63-year-old female patient presented with a large mass in the left mandibular alveolar ridge. Histologically, the lesion showed areas of conventional squamous cell carcinoma along with atypical epithelial cells forming a glandular pattern. However, there is no evidence of glandular differentiation, secretory activity or its products. Adenoid squamous cell carcinoma must be differentiated from adenosquamous carcinoma in which adenocarcinoma elements are positive for mucins. There are few cases reported to establish biological behaviour and prognosis.

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Introduction

Adenoid squamous cell carcinoma (ASCC) accounts for 2%-4% of all squamous cell carcinoma (SCC) cases. The sun-exposed areas of the skin, particularly on the head and neck of elderly men, are more commonly affected. [1] It was first described by Lever in 1947 as adenoacanthoma of the sweat glands. [2] Later Muller suggested the terminology of ASCC. ASCC has derived its name from the pseudoglandular appearance resulting from acantholysis and degeneration within the islands of SCC. [3] The first documented oral mucosal ASCC involving the tongue was reported by Goldman *et al.* in 1977. [4] ASCC arising in sun-exposed areas of skin seems to have a slightly greater risk of recurrence and metastasis than conventional SCC. [5] It has been suggested that intraoral ASCC are more aggressive with possible poor prognosis and clinicians should consider multidisciplinary treatment. [6] Prognosis of mucosal lesions are however controversial. We report a case arising in the oral mucosa proper, which is not exposed to sunlight and has no adnexal apparatus of the skin.

Case Report

A 63-year-old female patient reported with a swelling of the left lower jaw. (Figure 1)

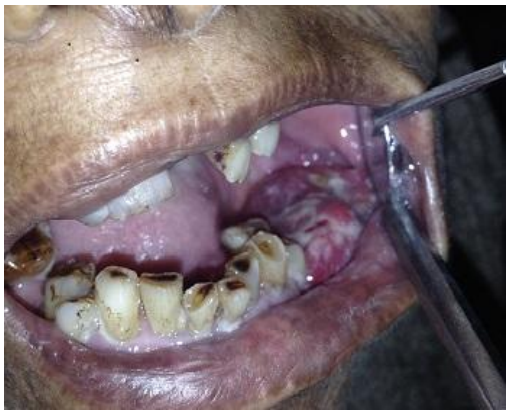

Figure 1: Clinical photograph showing ulceroproliferative growth in the mandibular left alveolus and buccal sulcular region

She noticed swelling about six months before, associated with pain radiating to the left temporal region. Patient had a habit of chewing betel quid 5-6 times a day for the past 30 years. On extra oral examination, there was facial asymmetry. Intra-orally an ulcerated, erythematous rough mass extending from tooth #34 to 38 region measuring about 3.5 x 2.5cm, covering buccal vestibule was seen. Panoramic radiography revealed a poorly defined radiolucency with ragged borders in relation to 35 and involving edentulous alveolar bone until molar ramus areas, extending inferiorly until mandibular canal. A soft tissue shadow is noted above the area involved. (Figure 2)

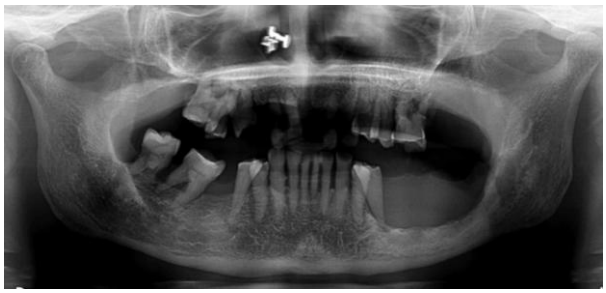

Figure 2: Panoramic radiograph showing ill-defined radiolucency with ragged borders extending from 34 until mandibular ramus area and a soft tissue shadow is also noted above the involved area

A solitary left submandibular lymph node was palpable, non-tender, and fixed to the underlying bone. On microscopic examination of the incisional biopsy the hematoxylin and eosin stained sections exhibited proliferation of dysplastic epithelium into connective tissue showing nuclear hyperchromatism, altered nuclear cytoplasmic ratio, individual cell keratinization, numerous normal, and abnormal mitotic figures. (Figure 3)

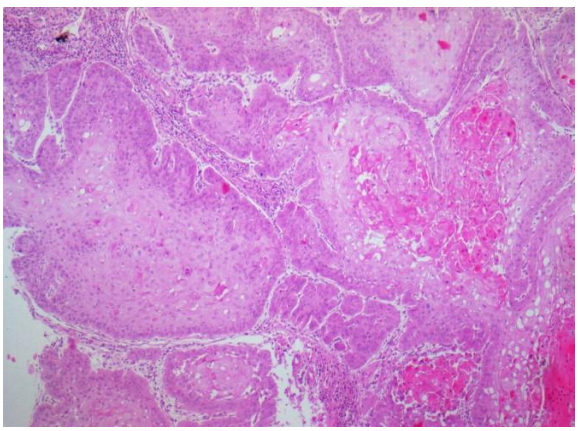

Figure 3: Hematoxylin and eosin stained section showing dysplastic epithelium infiltrating into the connective tissue in the form of islands and ductal pattern (10X).

Dysplastic epithelium infiltrating into the connective tissue were arranged in the form of islands and ductal pattern (Figure 4).

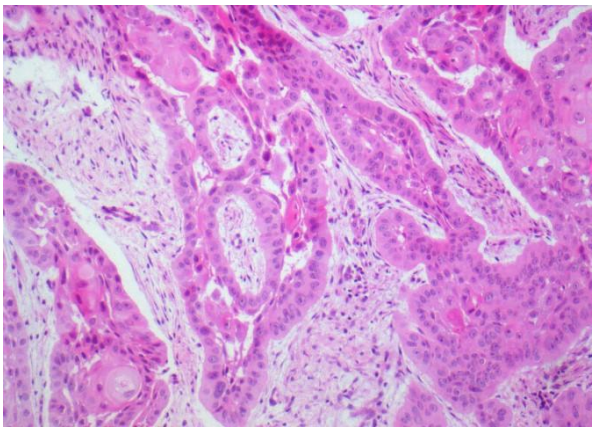

Figure 4: Histopathologic sections showing ductal pattern with peripherally lined columnar to cuboidal cells and central regions showing squamous cells, keratin pearls, and individual cell keratinization (20X).

These ductal patterns showed peripheral cells, which are of columnar to cuboidal with central regions showing squamous cells, keratin pearls and occasional individual cell keratinization (Figure 5).

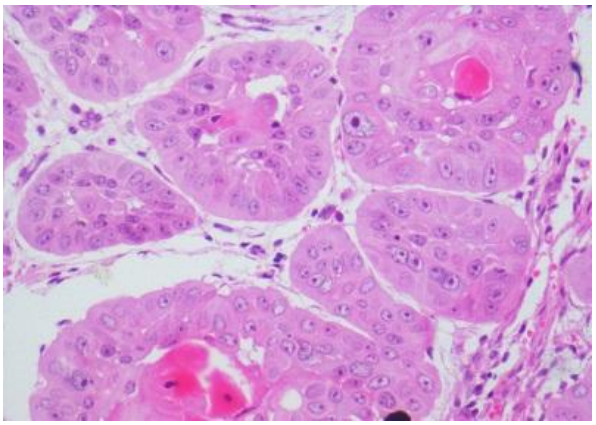

Figure 5: High power view of histopathologic section showing dysplastic epithelial cells arranged in ductal pattern (40X).

The surrounding connective tissue is made up of both mature and immature collagen fibers with moderate infiltration of chronic inflammatory cells. Periodic acid Schiff and Alcian blue staining showed no intracellular or extracellular mucinous material in the tumor, including acantholytic and pseudoglandular areas. These tumor cells were negative for mucicarmine, indicating that these cells are not glandular in origin and the possibility of mucoepidermoid carcinoma should be ruled out. (Figure 6) The diagnosis of ASCC was given. The

patient refused to undergo surgery, therefore, was referred to oncology institute for radiation therapy.

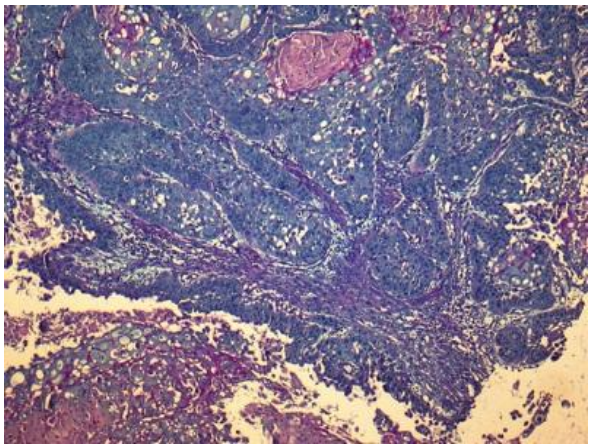

Figure 6: Histopathologic sections showing negativity for mucicarmine stain (10X).

Discussion

ASCC is an uncommon, histologic variant of SCC, microscopically characterized by nests of malignant epithelial cells revealing acantholysis in the centre of cancer islands leading to a pseudoglandular or pseudoluminal appearance. [1] Initially, the tumor was thought to arise from sweat glands. Later subsequent studies stated that the tumor is a variant of SCC of non-eccrine origin. [2] Muller *et al.* [3] reported additional seven cases and presented a review of literature with previously reported 15 cases. They emphasized on the unique histopathologic features, concurred that the gland like spaces are due to acantholysis in the solid nests of SCC, and suggested a terminology of adenoid SCC to avoid confusion with adenoacanthoma of endometrium. [3] Synonyms include adenoid SCC, pseudoglandular SCC, and SCC with gland like features, angiosarcoma-like SCC, and pseudovascular adenoid SCC. [5]

To our knowledge, 45 cases of oral ASCC (including lip) have been documented in English language literature including the present case. The cases reviewed from the literature are tabulated in Table 1.

Nearly half of the cases (22, 48.8%) reported to occur on lips (vermillion). Sixteen involving lower lip and 4 on upper lip and 2 not specified, rest 23 cases has been distributed in various regions of oral cavity, 8 cases involving gingiva, 6 affecting tongue, 6 floor of the mouth and 3 were found to present on buccal mucosa. Age ranges from 38-79 years with a mean age of 56.1 years. Thirteen female and 31 male patients with predilection towards male (70%) are observed in our review, consistent with Jacoway *et al.* [7] report with male predominance (87%).

The lesion on the lip are similar to that occur on skin exhibited as an elevated nodules that may show crusting, scaling or ulceration. [5] Most of the intra oral lesions appeared as exophytic polypoid ulceroproliferative growths. All the lesions on lip reported so far were measured less than 1.8 centimeters compared to larger (6cms) intra oral lesions. The intra-oral lesions are aggressive in nature and due to this behavior; they may not be identical to those of the skin and lip lesions. [5] The diagnostic criteria of ASCC [26] are basic cell of the keratinizing squamous cell type; adenoid structure consisting of a rounded space having a definite wall, principally of one cell thickness; lumen containing single or grouped dyskeratotic acantholytic cells. Our case fulfilled these strict criteria.

Oral ASCC should be differentiated from adenosquamous carcinoma, which is a rare aggressive variant of SCC that is characterized histopathologically by the combination of adeno carcinoma and SCC. The glandular pattern in ASCC includes mucous production that can be demonstrated clearly by mucicarmine stain. [27] The possibility of adenosquamous cell carcinoma was excluded, as histochemical examination was negative for mucin in the present case. Mucoepidermoid carcinoma of minor salivary glands may not show involvement of surface epithelium; evidence of mucous and intermediate cells can assist in diagnosis. In some instances, acantholysis may lead to formation of pseudovascular spaces, anastomosing channels resembling angiosarcoma; such variant of ASCC is called pseudovascular ASCC or angiosarcoma like SCC. Many a times, such tumors may be interpreted as angiosarcoma, in such instances immunohistochemical studies have shown that ASCC express cytokeratins and epithelial membrane antigens; angiosarcomas typically express vascular antigen markers like CD31, CD34 and Von Willebrand antigen. [14] Driemel *et al.* [17] emphasized on the immune histochemical analysis on three cases of ASCC and one case of angiosarcoma in order to differentiate these two tumors. Endothelial differentiating marker Fli-1 protein has been expressed in angiosarcomas and ASCC is immunonegative for this marker; instead, Laminin-5 is expressed in ASCC. So the authors

Table 1: A brief review of clinical features of the cases reported in the literature

Reference	No. of cases	Mean Age	Sex	Location	Size (in cm)	Lesion	Follow up
Jacoway *et al.* [7] 1971	15	56.1 (41- 75)	13M 2F	Lower lip(11) Upper Lip (3)	0.2-1.8	Tumor	NED [13] NS [2]
Tomich and Hutton [8] 1972	2	50 53	M M	Lower Lip(2**) Lower Lip	0.2-1.8	Ulcerated, keratotic indurated	NED [3]
Weitzner [9] 1974	1	67	M	Lower Lip	0.2-1.8		NED- DOC
Goldman [4] 1977	1	61	M	Posteriolateral part of tongue	NS	Ulcerated tumor	DOD [8]
Takagi *et al.* [10] 1977	2	50 56	F M	Maxillary Gingiva Lateral part of Tongue	NS	Ulcerated tumor erosions	DOD RE [24] DOD RE [20]
Caya *et al.* [11] 1985	1	50	M	Lip	NS	NS	NS
Sivapathasundaram and Roshini [5] 1992	1	NS	NS	Gingiva	NS	NS	DOD
Jones *et al.* [12] 1993	3	58 47 42	M M F	Floor of the mouth Lower Lip(2)	2 X 1(2) 1X1	Verrucous exophyt-ic, growth	NED [2] NS
Blackburn et al[13] 1999	1	78	F	Upper Lip	1X1	NA	NED
Zidar *et al.* [14] 2006	2	59 77	M F	Buccal Mucosa Floor of mouth	2 x 2x0.5 4x2x2	Ulcerated lesion with indurated, slightly raised mar-gins polypoid tumor	NED [20] NED [16]
Kasafuka *et al.* [16] 2006	1	64	F	Floor of mouth	2x1	Tumor	NED [5]
Driemel *et al.* [17] 2008		57 68 50	M M M	Tongue Floor of mouth	NA NA NA	NA NA NA	NA NA NA
Kerawala *et al.* [5] 2009	1	56	M	Lateral side of Tongue	1.6x1.1	Ulcer	RE [5] DOD [4]
Papadopoulou *et al.* [18] 2010	1	72	F	Mandibular alveolar ridge	1.7 x 1	Irregular mass, with a central ulceration	DOD [7] RE [10]
Prasad *et al.* [19] 2010	1	70	F	Gingiva	NA	NA	NA
Yeoh MS *et al.* [20] 2012	1	38	F	Buccal mucosa	3.2x5	Ulceroproliferative growth	DOD [7] RE [6]
Terado *et al.* [21] 2012	1	73	F	Mandibular alveolar ridge	1.5x1.5x1	Granulation tissue	NED
Nayak *et al.* [22] 2012	2	45 53	M M	Floor of mouth Maxillary alveolar ridge	3.5 x 2.5 x 2 2 x 5.5 x 1.5	Erythematous in-flamed mass Linear proliferative growth	NED NED
Vidyavathi K. [15] 2012	1	40	M	Floor of mouth	6x6x3	Polypoid growth	NS
Patil SK *et al.* [23] 2014	1	49	F	Buccal mucosa	3.5x2	Ulceroproliferative growth	NED
Deepak *et al.* [24] 2014	1	38	M	Tongue	2x2	Ulceroproliferative growth	NS
Kavita Mardi *et al.* [25] 2014	1	50	M	NA	3x3	Ulcerative lesion	NS

NED- no evidence of disease, NS – not specified, NA- not available, RE-recurrence, DOD- died of disease, DOC- death due to other cause.**-two lesions

recommended Fli-1 as a discriminating factor to differentiate these two tumors.

Laminin-5 also acts as a tumor biological indicator of the unfavorable prognosis of ASCC. [17] Loss of expression of adhesion molecule E- cadherin has been observed in ASCC. Studies have shown that E- cadherin is expressed in most SCC of the head and neck, the expression being strong in well-differentiated cancers, but reduced in poorly differentiated tumors. [14]

Though the biological behavior and prognosis is

debatable, the cases reported so far on lip (vermillion) has good prognosis with no evidence of metastasis and recurrence or death due to disease. This can be attributed to their location, easily recognizable site and seek treatment at the earliest and small lesions resolves after local excision. Six cases recurred in a period of 5-24 months, seven cases died of disease, one case died with other cause, and eight cases are free of disease. According to some authors, intraoral ASCC has aggressive clinical nature with poor prognosis, [5-6, 18, 20] and to prompt clinicians to consider a more aggressive multidisciplinary treatment for these cases. [5] At the same time, there are some intra mucosal cases (8 cases) with no evidence of recurrence. [12-14, 16, 21-23]

The current study had some limitations since immunohistochemistry and other advanced diagnostic aids were not used in this study to confirm the cell of origin of the lesion. Moreover, excisional biopsy was not obtained as the patient refused to undergo surgical treatment.

Conclusion

ASCC of the oral cavity is very rare. Oral ASCC may show pseudoglandular and pseudovascular morphology. ASCC must be differentiated from adenosquamous carcinoma in which adenocarcinoma element is positive for mucins. Prognosis of the mucosal lesions is controversial, because the cases of intra oral ASCC reported are too small to elucidate the biological behavior and prognosis.

Conflicts of Interest

None to declare.

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