Case Report

Adenoid Squamous Cell Carcinoma on Buccal Mucosa: A Case Report


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Abstract:
Squamous cell carcinoma (SCC) is the most common malignancy arising within the oral cavity. The adenoid/acantholytic squamous cell carcinoma (ASCC) is an uncommon but a well-recognized variant. In the head and neck region, ASCC has been reported mostly on the sun exposed regions of the skin with only around 34 cases being reported in the oral cavity. Histologically, this variant presents with areas of conventional SCC intermixed with atypical cells exhibiting pseudoglandular or an adenoid arrangement. The clinical behavior of ASCC on the skin is considered to be more aggressive than the conventional SCC but there is insufficient data regarding those arising within the oral cavity. Here we report a case of ASCC occurring on the buccal mucosa by far the rare case reported in this site.

Key words: Acantholytic squamous cell carcinoma, Adenoid squamous cell carcinoma, Pseudoglandular, Adenoacanthoma.

Introduction
Squamous cell carcinoma (SCC) is considered to be the most common malignancy arising within the oral cavity. Histological variants such as Verrucous carcinoma, Spindle cell carcinoma, Basaloid SCC, Adenosquamous carcinoma, Adenoid SCC (ASCC) and undifferentiated SCC have been reported in the literature.1-2 The microscopic features of most of these variants overlap considerably with those of other lesions such as salivary gland neoplasms, connective tissue tumors, odontogenic lesions and verrucopapillary pathologies thus proving to be a challenge for the histopathologist.

An interesting variant of SCC with pseudoglandular spaces or lumina has been reported frequently on the sun exposed regions of the skin.3 Till date a total of thirty four cases of this adenoid or pseudoglandular variant has been reported in the oral cavity a majority of which occurred on the lip followed by gingiva, floor of the mouth and maxillary ridge.4 ASCC arising on the skin is considered to be more aggressive than the conventional SCC. Some authors consider the intraoral ASCC to be far more aggressive than ASCC of skin but this fact cannot be substantiated due to the paucity of reported cases.3,4 Here we report a case of adenoid
squamous cell carcinoma of buccal mucosa, which is by the rare case reporting on this site.

Case Report

A 49 year old female patient reported to our institution, with a chief complaint of pain in left cheek region since 1 month. She had the habit of chewing tobacco and alcohol consumption for the past 20 years. On extra oral examination a sinus opening with pus discharge on the left cheek was noted. The surrounding skin was tender on palpation, firm and fixed to the underlying structures. Left submandibular lymph node was palpable measuring 1x1 cm, tender, firm and mobile.

On intraoral examination, an ulceroproliferative growth measuring 3.5 X 2.0 cm was seen on the left buccal mucosa, extending from mandibular first premolar to the retromolar trigone anteroposteriorly and from the upper vestibule to the lower vestibule superoinferiorly. The lesion was tender on palpation with indurated borders. Orthopantomogram and computed tomography revealed no bony involvement of either the maxilla or the mandible. Based on the history and clinical findings a provisional diagnosis of malignancy was made. An incisional biopsy was performed. A final diagnosis of conventional, well differentiated squamous cell carcinoma was made after routine processing and staining of the specimen with Haematoxylin and Eosin. Excision of the lesion along with modified radical neck dissection was planned and performed. Histopathology of the excised specimen, revealed the presence of epithelium and connective tissue. The epithelium was parakeratinised stratified squamous exhibiting dysplastic features like loss of basal cell polarity, basal cell hyperplasia, loss of stratification, increased nuclear and cytoplasmic ratio, nuclear and cellular pleomorphism, few abnormal mitotic figures.
Figure 4: Alcian blue PAS stain showed negative for adenoid squamous cell carcinoma and individual cell keratinization. Connective tissue shows tumor islands with central area of detached neoplastic or acantholytic neoplastic cells lined by polygonal cells of approximately 2 to 3 cells in thickness. These areas gave an adenoid/pseudoglandular like appearance. (Figure 1 and 2) Stroma is also intermixed with areas of conventional SCC. Stains for mucin (mucicarmine and alcian blue) turned out to be negative. (Figure 3 and 4) Thus a diagnosis of adenoid squamous cell carcinoma was made. The patient was referred to an oncology center for radiotherapy. Currently the patient is under constant follow up.

Discussion
ASCC was first described in 1947 by Lever as an adenoacanthoma, assuming it to arise from the eccrine sweat ducts and glands. Later, Lever modified this concept and stated that the gland like spaces was the result of acantholysis of solid nests of squamous cell carcinoma. Muller and colleagues suggested the term Adenoid squamous cell carcinoma (ASCC). Other synonyms include pseudoglandular SCC and Acantholytic SCC.² Clinically, ASCC occurs predominantly in male patients during the 6th decade or older.¹ Most commonly this variant has been reported on the sun exposed regions of the skin. Lesion on the skin appears as an elevated nodule that may show crusting, scaling and ulceration. The borders of the lesion exhibit an elevated or a rolled out appearance.³ Microscopically ASCC shows duct like spaces intermixed with areas of conventional SCC. These duct-like structures are lined by a single or double layer of cuboidal or polygonal epithelial cells often containing dyskeratotic and acantholytic cells in the lumen. Connective tissue stroma demonstrates variable collagen fibres and inflammatory infiltrate.¹,³ Histochemical analysis of mucin with mucicarmine and alcian blue stains are negative.¹ Immunohistochemistry of these tumors demonstrates positivity for cytokeratins (AE1/AE3) and epithelial membrane antigen. Factor VIII-related antigen, S-100, Carcinoembryogenic antigen, CD 34 are negative.¹,² Ultrastructural findings have supported squamous origin (presence of desmosomes and hemidesmosomes) and no glandular features such as intracytoplasmic microvilli and secretory granules.² The histopathology of the present case is consistent with the above mentioned findings in ASCC.

The differential diagnosis includes adenosquamous carcinoma, mucoepidermoid carcinoma, adenoid cystic carcinoma.¹,⁵ Adenosquamous carcinoma is a rare dimorphic neoplasm which possess histopathological features of adenocarcinoma and SCC. The glandular elements in this neoplasm positive for mucin stain which helps to differentiate this variant from Adenosquamous carcinoma. The lack of myoepithelial cells and mucous cells helps to rule out adenoid cystic carcinoma and mucoepidermoid carcinoma respectively.²,⁵ Pseudovascular ASCC a rare variant of ASCC needs to be distinguished from angiosarcoma.⁶ CD34, CD31, factor VIII-related antigen are positive for angiosarcoma. Some rare benign disorders have to be
differentiated also, like acantholytic seborrhoic keratosis and isolated dyskeratotic acanthoma which may be histologically similar. Careful examination reveals the lack of dysplastic features in these conditions.  

**Conclusion:**
ASCC of skin has been noted to be more aggressive than conventional SCC but how far this is true within the oral cavity needs further evaluation. The dearth of adequate reporting and data on these variants has hampered further research. The histopathologist has to be aware of this void and identify these variants to help shed light on these lesions. Also further studies are required to analyze whether the recognition of these variants in SCC affect the treatment plan and prognosis. If proved, should these histopathological variations be included as criteria's in staging of the lesions – a yet unanswered question.

**References:**

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