

Adenoid squamous cell carcinoma of upper lip, a rare variant of squamous cell carcinoma: A case report

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ABSTRACT

Adenoid squamous cell carcinoma (ASCC) is a variant of squamous cell carcinoma. As this variant is rare it is difficult to distinguish it from other variants of squamous cell carcinoma. It commonly originates in the head and neck region as these parts are highly exposed to the sunlight. There are very few cases reported in the literature. Here we report a case in a 30-year-old male who reported with a nodular lesion over the upper lip with no unusual signs, which was confirmed as adenoid squamous cell carcinoma after various histopathological tests.

Keywords: Adenoid squamous cell carcinoma, nodular, squamous cell carcinoma, vermillion

Introduction

Malignant lesions on the upper lip are uncommon when compared to those which are seen on lower lip.^[1] Adenoid squamous cell carcinoma (ASCC) is a rare entity of squamous cell carcinoma of which only 45 cases have been reported in the literature.^[2] The number of cases reported in the past has not allowed conclusive characterization of ASCC of the lip. However, those lesions previously described have affected predominantly males.^[3]

Case Report

A 30-year-old male reported with a nodule with raised margins, which was dome-shaped, painless, black, originating from the

sub-mucosa of the middle part of the upper lip [Figure 1]. On eliciting a history, the patient had a forceful impact on the upper lip by a branch of a tree while farming 2 years back. The patient was a habitual smoker for the past 7 years with no relevant medical history. On inspection, the lesion was well-circumscribed and measured 1 cm in diameter with slight surface ulceration. On palpation, it was hard in consistency. It was highly suspected to be a carcinoma considering its ulcero-proliferative characteristics and thus was planned for excisional biopsy.

Histopathological reports revealed both squamous and glandular differentiation. Individual cell keratinization and keratin pearl formation was evident along with few areas of abundant islands exhibiting areas of cystic degeneration, representative of pseudo glandular differentiation [Figure 2]. Keratin pearl formation was indicative of squamous cell carcinoma and glandular differentiation suggestive of Adenoid squamous cell carcinoma [Figure 3].

ABBREVIATION: C.S: Cross-section; L.S: Longitudinal section; OSCC: oral squamous cell carcinoma; ASCC: adenoid squamous cell carcinoma

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Discussion

ASCC is uncommon, but well-described, a variant of squamous cell carcinoma. ASCC occurs most commonly on the skin of the head and neck. Cutaneous ASCC has been reported to recur, metastasize, and has been associated with other synchronously occurring malignant neoplasms.^[3]

ASCC has been considered a rare entity containing pseudo-glandular spaces or lamina histologically.^[4] There are very minimal reported cases of ASCC in literature until 1970, seventeen patients with adenoid squamous cell carcinoma on the vermilion mucosa of the lip were documented. The first intra oral case of ASCC was reported by Goldman in 1977.^[5]

Several other authors^[2] conducted a study comprising of a total number of 45 cases of ASCC, of which 16 involved lower lip, 4 on the upper lip, 2 were not specified and rest 23 cases were distributed in various regions of the oral cavity as tabulated in Table 1. In contrary to normal oral squamous cell carcinoma (OSCC) incidence, the lesion was seen on the upper lip in our case.

Few authors^[6] stated that the age ranged between 38-79 years with a mean age of 56.1 years, stating middle and elderly aged groups are more susceptible. Male patients showed more

predilection (70%) population, which was in accordance with our report. We also emphasize that, its apparent origin is from the underlying traumatically damaged tissue in addition to smoking, sun exposure during farming.

ASCC share many common histological features with other variants of SCC but the presence of squamous and glandular structures histologically separates it from ASCC as seen in the figure above.^[7]

A lesion on the vermilion border makes it difficult to determine whether it is originating from the vermilion, the underlying mucosa, or from the overlying skin. Based on the history and the site of the lesion, we suspected lesion to be either a traumatic ulcer, cyst (retention/extravasation cyst) or tumor arising from the skin appendages, salivary glands, or mesenchymal origin.

Cystic lesions can be differentiated from solid tumors by palpation. However, the presence of secondary inflammation and the formation of granulation tissue in the cystic structure makes it difficult to identify when they are ruptured^[8] which made us consider the above-mentioned diagnosis.

Differential diagnosis of the lesion could be keratoacanthomas was completely ruled out, as 8% occur on the upper lip and they



Figure 1: Clinical photograph showing black submucosal nodular growth on the midline of vermilion border of the upper lip

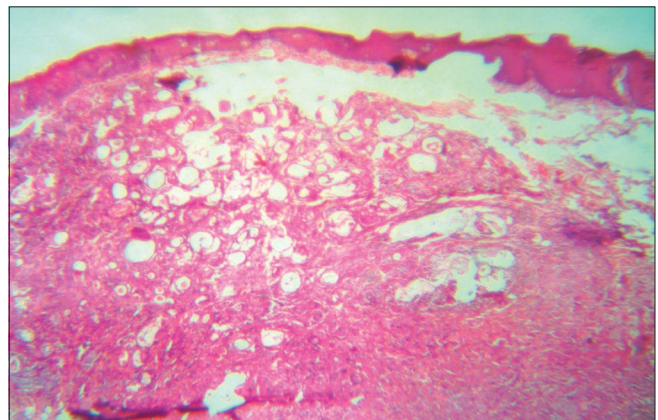


Figure 2: Haematoxylin and eosin stained section exhibiting both squamous and glandular differentiation and showing dysplastic epithelium arranged in small islands

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

REFERENCE	NO OF CASES	MEAN AGE	SEX	LOCATION	SIZE	LESION	FOLLOW UP
Jacoway <i>et al.</i> ^[6] 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 ^[13] 1972	2	50 56	M M	Lower lip(2**) Lower lip	0.2-1.8	Ulcerated, keratotic indurated	NED [3]
Weitzner ^[14] 1974	1	67	M	Lower lip	0.2-1.8		NED-DOC
Caya <i>et al.</i> ^[15] 1985	1	50	M	Lip	NS	NS	NS
Jones <i>et al.</i> ^[6] 1993	2	47 42	M F	Lower lip Lower lip	1x1		NED-2
Blackburn <i>et al.</i> ^[3] 1999	1	78	F	Upper lip	1x1	NA	NED

Abbreviations: NED- no evidence of disease, NS- not specified, NA- not available, DOC- death due to other cause,**two primary lesions of lower lip 14 months apart

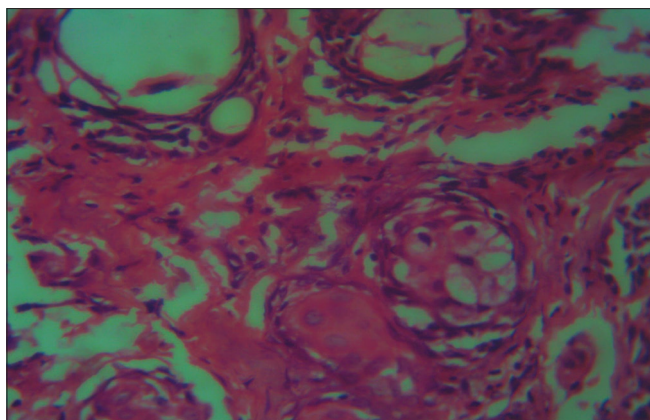


Figure 3: Histopathological section showing individual cell keratinization and keratin pearl formation

typically exhibit as central keratin filled depression.^[9] Likewise, squamous cell papilloma, haemangiomas, nodular lesions like a lipoma or oral leiomyoma can also occur on lips. None of the above findings were in common with our lesion. Absence of infection and lip-biting habit ruled out acute periapical abscess of maxillary anterior teeth or fibrous hyperplasia caused by trauma such as lip biting may also result in swelling.^[10]

Diagnostic criteria for ASCC histologically are the presence of basal cells 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.^[11] In our present case, the squamous component was predominant, moderately to poorly differentiated and showed prominent dyskeratosis, there was lack of perfusion of nutrition to the acantholytic cells from adjacent connective tissue stroma.

For differential diagnosis immunohistochemical characteristics are very helpful, such as PASCC expresses epithelial markers like cytokeratin and epithelial membrane antigen on the immunohistochemical study.^[12] Therefore, to get a histopathological differential diagnosis and rule out the similar clinically appearing lesions immunohistochemical characteristics should be performed.

Few authors^[3] reported a case of primary ASCC of the upper lip associated with a locoregional metastasis. Our case had no such significant findings but kept under observation.

Few authors reported intraoral recurrence and had to be treated with chemotherapy.^[5] Others stated a secondary lesion occurred on the lower lip.^[13] In our case report, there were no lesions seen intraorally making the lesion as a primary lesion.

The patient has been under our observation for the past 15 months with no recurrence observed [Figure 4]. Therefore, a simple clinical presentation may be misleading and may result in a casual attitude towards the lesion. So, precise histopathological diagnosis can help the clinician to plan accurate treatment.

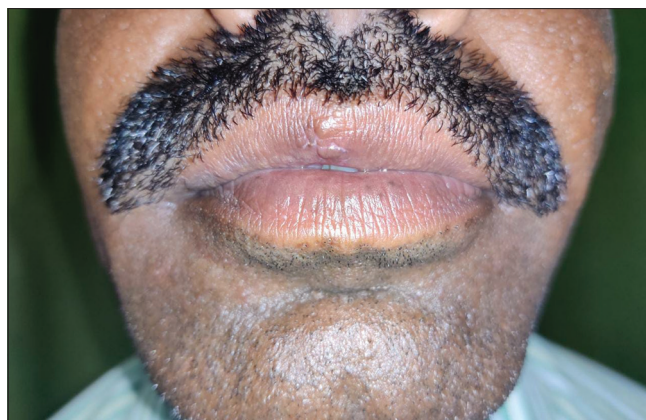


Figure 4: Clinical photograph on 6 months follow up period

Conclusion

ASCC of the oral cavity is very rare, it shows pseudo-glandular and pseudo-vascular 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 intraoral ASCC reported are too small to elucidate the biological behavior and prognosis.

Ethics statement/confirmation of patient's permission

Informed consent was obtained for experimentation with human subjects.

Ethical and institutional permission were taken.

Key points

- Site of the lesion
- Rate of occurrence
- Prognosis in controversial Precise histopathological diagnosis can help to evaluate and plan for treatment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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