

Adenoid squamous cell carcinoma of buccal mucosa – A rare case report

ABSTRACT

Adenoid squamous cell carcinoma is a rare variant of squamous cell carcinoma. It is mainly reported in the sun-exposed areas of the head and neck. In literature, adenoid variants of squamous cell carcinoma are reported in the lip and tongue. Documented cases of buccal mucosa are very few. In this case report, we describe an adenoid variant of squamous cell carcinoma, which is an unusual presentation of a rare carcinoma.

Keywords: Adenoid, carcinoma, cavity, cell, oral, squamous

INTRODUCTION

Adenoid squamous cell carcinoma (ASCC) is a rare variant of squamous cell carcinoma. It accounts for about 2–4% reported cases of squamous cell carcinoma.^[1,2] It is widely accepted as an uncommon, distinctive histological variant. It was first documented in literature by Lever in sweat glands and is called adeno acanthoma.^[1] Microscopically, they show an adenoid pattern and hence are called pseudoglandular squamous cell carcinoma. Owing to its glandular pattern, Muller^[3] introduced the term Adenoid squamous cell carcinoma. This condition is often observed in sun-exposed areas of the head and neck.^[4] It was first considered to develop from the sweat glands, but later on, cases on the oral cavity and oropharynx that are not exposed to sunlight and have no adnexal apparatus of the skin are documented.^[1] Adenoid squamous cell carcinoma variety shows more recurrence and metastatic potential than the classical variety of squamous cell carcinoma (SCC).

CASE REPORT

A 61-year-old gentleman reported to our outpatient department with the chief complaint of non-healing ulceration in the right and left side of his mouth for 9 months. Three months after ulceration developed on the

left side too. The ulceration was first noted on the right side. Patient had deleterious habit of cigarette smoking for twenty years with occasional alcoholism. Medical history was unremarkable and the review of his systems appeared normal.


On examination ulcero-proliferative erythematous lesion of 1.5 cm × 1 cm was present on the right buccal mucosa near the retromolar area. The lesion showed peripheral induration and was tender on palpation. There was also pain during mouth opening on the right side. The left side buccal mucosa near the vestibular area of 27, 28 area showed a heterogeneous white lesion with central ulceration of size 0.5 cm × 0.5 cm [Figures 1 and 2].

AKHILESH PRATHAP, NIDHIN RAJAGOPALAN SREEJA, SUNIL SUKUMARAN NAIR¹, SHARLENE S. BABU¹

Departments of Oral and Maxillofacial Surgery and ¹Oral Pathology, Pushpagiri College of Dental Sciences, Kerala University of Health Sciences, Thiruvalla, Kerala, India

Address for correspondence: Dr. Nidhin Rajagopalan Sreeja, Assistant Professor, Department of Oral and Maxillofacial Surgery, Sri Sankara Dental College, Varkala, Trivandrum, Kerala, India. E-mail: nithinrajagopalan@gmail.com

Received: 03 September 2022, **Revised:** 25 November 2022, **Accepted:** 19 May 2023, **Published:** 10 November 2023

Access this article online	
Website: www.njms.in	Quick Response Code 
DOI: 10.4103/njms.njms_158_22	

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Prathap A, Sreeja NR, Nair SS, Babu SS. Adenoid squamous cell carcinoma of buccal mucosa – A rare case report. Natl J Maxillofac Surg 2023;14:492-5.

Investigations performed

A complete blood examination was done and all the parameters were within normal limits.

Procedure done

Excisional biopsy was performed for the lesion in relation to the left buccal mucosa and incisional biopsy was performed on the right side and both samples were fixed in 10% formalin and were sent for histopathological examination.

Histopathological report

(1) Right side

The section showed non-keratinized stratified squamous epithelium showing suprabasilar clefting with acantholytic cells. The connective tissue showed invading epithelium in the form of islands, sheets, and adenoid pattern is noticed. The underlying connective tissue was moderately collagenous and vascular. The microscopic findings were suggestive of adenoid squamous cell carcinoma [Figures 3 and 4].

(2) Left side

The sections showed connective tissue, which was moderately collagenous and vascular. An overlying epithelium was not seen. The connective tissue showed the invading epithelium in the form of islands. The margins showed adequate clearance from the tumor. The histopathologic findings were suggestive of squamous cell carcinoma.

Referral

The patient was referred to our oncosurgery department for further management. But unfortunately, due to the financial constraints, patient was not willing for further treatment and the follow-up was lost.

DISCUSSION

Adenoid squamous cell carcinoma is an uncommon, distinctive histological variant of squamous cell carcinoma.^[5] Due to its pseudoglandular or pseudoluminal appearance, it is called adenoid squamous cell carcinoma. Even though it's a rare malignancy, about 45 cases are reported in the available literature [Table 1]^[1] (excluding our case). Some of the important documented case reports are listed in Table 1. Of these documented cases, majority of the cases are reported in the lower lip. Reported cases in buccal mucosa are very few.^[1] This makes our case report exceptional.

It was originally described by Lever in 1947. This variant is produced because of acantholysis and degeneration within islands of squamous cell carcinoma. The result is a pseudoadenocarcinomatous appearance, but there is no evidence of glandular differentiation or of secretory activity or products.^[18]



Figure 1: Site of lesion in right buccal mucosa near the retromolar area



Figure 2: Site of lesion in left buccal mucosa

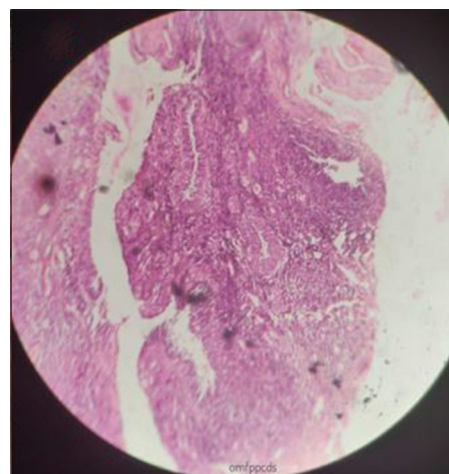


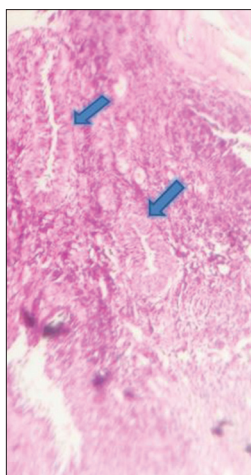
Figure 3: Histopathological image

Clinical features

Adenoid squamous cell carcinoma is reported to occur as early as 20 years of age. But according to documented evidence by Johnson and Helwig, nearly 75% patients were 50 years of age

Table 1: Reported cases of adenoid variety of squamous cell carcinoma

Reference	Year	Site	Number of cases reported
Gerughty <i>et al.</i> ^[6]	1968	Nasal, oral, and laryngeal Cavities	10 cases
Jacoway <i>et al.</i> ^[7]	1971	Oral labial mucosa	15 cases
Lasser <i>et al.</i> ^[8]	1974	Uvula	1 case
Goldman <i>et al.</i> ^[9]	1977	First reported case of ASCC in tongue	1 case
Takagi <i>et al.</i> ^[10]	1977	Maxillary Gingiva Lateral part of Tongue	2 cases
Caya <i>et al.</i> ^[11]	1985	Lip	1 case
Jones <i>et al.</i> ^[12]	1993	Floor of the mouth Lower Lip	2 cases
Blackburn <i>et al.</i> ^[13]	1999	Upper lip	1 case
Zidar <i>et al.</i> ^[14]	2006	Buccal Mucosa* Floor of mouth	2 cases
Driemel <i>et al.</i> ^[15]	2008	Tongue Floor of mouth	2 cases
Yeoh <i>et al.</i> ^[16]	2012	Buccal Mucosa*	1 case
Donthi <i>et al.</i> ^[17]	2014	Tongue	1 case
Chandrakala <i>et al.</i> ^[1]	2018	Mandibular alveolar ridge	1 case

**Figure 4: Higher magnification image of histopathological section**

or older. In their series, only 98% patients were males. Hence, ASCC shows obvious male predilection.^[19] They are mainly seen in the sun-exposed areas of the head and neck (93%).^[19] The lesions on the skin appear as simply elevated nodules that may show crusting, scaling, or ulceration. Sometimes, there is an elevated or rolled border to the lesion. Intraoral lesions are rare. Sivapathasundharam B and Rohini S are credited to report the first case from India.^[18] They reported four cases that occurred in gingiva. Out of these four cases, three of these lesions behaved in a very aggressive fashion, metastasized in at least two instances and the patient died in all three cases because of the tumor. Because of the aggressive nature of the lesions, these intraoral cases may not be identical to those of the skin and lips.

Histologic features

There is a proliferation of surface dysplastic epithelium into the connective tissue as in a typical epidermoid carcinoma.

However, the lateral or deep extensions of this epithelium show the characteristic solid and tubular ductal structures, which typify the lesion. These duct-like structures are lined by a layer of cuboidal cells and often contain or enclose acantholytic or dyskeratotic cells.

Treatment and prognosis

Adenoid squamous cell carcinoma is generally treated by surgical excision. On only rare occasions does it metastasize or cause death of the patient. The biological behavior and prognosis of ASCC are debatable. Recurrence is relatively common (38% in the series of lip lesions reported by Jacoway and his coworkers,^[7] although it is possible that some of these may have been the second lesions, since multiple adenoid squamous cell carcinomas in the same patient often occur).^[7] According to Chandrakala and associates, the cases reported so far on lips (vermillion) have a good prognosis with no evidence of metastasis and recurrence. This can be correlated with their location, easily identifiable area, which helps to diagnose the disease at the earliest so that treatment can be done immediately. However, according to Sivapathasundharam B and Rohini S, cases reported in gingival showed aggressive behavior and obvious malignant potential. According to Driemel and coworkers,^[15] laminin-5 is a biological indicator of the unfavorable prognosis of ASCC.

Differential diagnosis

Adenoid squamous cell carcinomas should be distinguished from certain salivary neoplasms that may occur on the floor of the tongue, particularly adenoid cystic carcinomas (cylindroma) and mucoepidermoid carcinomas.^[20] The lesion resembles the former only semantically; although cutaneous examples of adenoid squamous carcinoma may contain a small amount of intraluminal hyaluronic acid, the

presence of vacuolated, mucin-producing cells and true glandular structures in mucoepidermoid carcinoma should help differentiate it from adenoid squamous cell carcinoma. Oral ASCC must also be differentiated from adenosquamous carcinoma, which is a rare combination of adeno-carcinoma and SCC. Adenoid squamous cell carcinoma produces luminal mucous that can be stained with mucicarmine.^[21] Angiosarcoma-like SCCs are also similar to ASCC histologically. Immune histochemical analysis angiosarcoma-like SCC expresses Fli-1 protein, which is an endothelial-differentiating marker. Fli-1 protein is negative in ASCC14.

CONCLUSION

Adenoid squamous cell carcinoma is a rare variant of squamous cell carcinoma with features of adenoid pattern. Since there are only a few reported cases, the exact biological behavior and prognosis of this variant is not conclusive. However, most of the pathologists agree that ASCC variety shows more recurrence and metastatic potential than the classical variety of SCC. Hence, aggressive treatment and regular follow-up is mandatory.

Declaration of patient consent

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

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Chandrakala J, Srinath S, Giraddi GB, Kendole RK. Adenoid squamous cell carcinoma of oral cavity: A case report. *J Dent Shiraz Univ Med Sci* 2018;19:68-73.
- Lever WF. Adenoacanthoma of sweat glands; carcinoma of sweat glands with glandular and epidermal elements: Report of four cases. *Arch Derm Syphilol* 1947;56:157-71.
- Muller SA, Wilhelmj CM Jr, Harrison EG Jr, Winkelmann RK. Adenoid squamous cell carcinoma (adenoacanthoma of lever). Report of seven cases and review. *Arch Dermatol* 1964;89:589-97.
- Cardesa A, Zidar N. Adenoid squamous cell carcinoma. In: Barnes L, Eveson JW, Reichart PA, Sidransky D, editors. WHO Classification of Tumours. Pathology and Genetics. Tumor of the Head and Neck. 1st ed. Lyon: IARC Press; 2005. p. 129.
- Tomich CE, Hutton CE. Adenoid squamous cell carcinoma of the lip: Report of cases. *J Oral Surg* 1972;30:592-8.
- Gerughty RM, Hennigar GR, and Brown FM. Adenosquamous carcinoma of the nasal, oral and laryngeal Cavities - A clinicopathologic survey of ten cases. *Cancer* 1968;22:1140-55.
- Jacoway JR, Nelson JF, Boyers RC. Adenoid squamous cell carcinoma (adenoacanthoma) of the oral labial mucosa-A clinicopathological study of 15 cases. *Oral Surg* 1971;32:444-9.
- Lasser A, Cornog JL, Morris JM. Adenoid squamous cell carcinoma of the vulva. *Cancer* 1974;33:224-7.
- Goldman RL, Klein HZ, Sung M. Adenoid squamous cell carcinoma of the oral cavity. *Arch Otolaryngol* 1977;103:496-8.
- Takagi M, Sakota Y, Takayama S, Ishikawa G. Adenoid squamous cell carcinoma of the oral mucosa: Report of two autopsy cases. *Cancer* 1977;40:2250-5.
- Caya JG, Hidayat AA, Weiner JM. A clinicopathologic study of 21 cases of adenoid squamous cell carcinoma of the eyelid and periorbital region. *Am J Ophthalmol* 1985;99:291-7.
- Jones AC, Freedman PD, Kerpel SM. Oral adenoid squamous cell carcinoma: A report of three cases and review of the literature. *J Oral Maxillofac Surg* 1993;51:676-81.
- Blackburn TK, Macpherson D, Conroy B. Primary adenoid squamous cell carcinoma of the upper lip associated with a locoregional metastasis: A case report and review of the literature. *J Oral Maxillofac Surg* 1999;57:612-6.
- Zidar N, Gale N, Zuperc A, Dovsak D. Pseudovascular adenoid squamous-cell carcinoma of the oral cavity -- A report of two cases. *J Clin Pathol* 2006;59:1206-8.
- Driemel O, Müller-Richter UD, Hakim SG, Bauer R, Berndt A, Kleinheinz J, et al. Oral acantholytic squamous cell carcinoma shares clinical and histological features with angiosarcoma. *Head Face Med* 2008;4:17. doi: 10.1186/1746-160X-4-17.
- Yeoh MS, Kim DD, Ghali GE. Acantholytic squamous cell carcinoma of the buccal mucosa: Report of a case. *J Oral Maxillofac Surg* 2012;70:1733-8.
- Donthi D, Ramaswamy AS, Mahalingasetti PB. Acantholytic squamous cell carcinoma of the tongue: A diagnostic challenge. *Clin Cancer Investig J* 2014;3:179-81.
- Rajendran R. Benign and malignant tumors of the oral cavity. In: Rajendran R, Shivapathasundaram B, editors. Shafer's Textbook of Oral Pathology. 7th ed. New Delhi, India: Elsevier India; 2012. p. 124.
- Johnson WC, Helwig EB. Adenoid squamous cell carcinoma (adenoacanthoma). *Cancer* 1966;19:1939.
- Goepfert H, Giraldo AA, Byers RM, Luna MA. Salivary gland tumors of the base of the tongue. *Arch Otolaryngol* 1976;102:391-5.
- Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and Maxillofacial Pathology. 2th ed. Philadelphia: Saunders; 2006. p. 370.