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Case Report

Adenoid cystic carcinoma of the buccal mucosa: A case report and review of the literature

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ABSTRACT

Adenoid cystic carcinomas are deceptive malignancies that show slow growth and local invasion with recurrences seen many years after diagnosis. Upto 50% of these tumors occur in the intraoral minor salivary glands usually in the hard palate. Buccal mucosal tumors are relatively rare. We determined the incidence of buccal mucosal adenoid cystic carcinoma by reviewing the number of reported cases in the literature. This is the first article to analyze the occurrence of adenoid cystic carcinomas in the buccal mucosa through a review of 41 articles. Our review revealed 178 buccal mucosal adenoid cystic carcinomas among a total of 2,280 reported cases. We present a case of adenoid cystic carcinoma occurring in the left buccal mucosa of a 45-year-old female.

Key Words: Adenoid cystic, carcinoma, salivary glands, ultrasonography

INTRODUCTION

Adenoid cystic carcinoma (ACC) is a malignant neoplasm of the salivary glands. It was first described as cylindroma by Billroth in 1856.[1] The term 'adenoid cystic carcinoma' was coined in the year 1928 and is in use till date.[2] Adenoid cystic carcinomas constitute less than 1% of all head and neck malignancies with 50% of all ACCs occurring intraorally, commonly in the hard palate.[2,3] Other less common intraoral sites include the lower lip, retromolar/tonsillar pillar region, sublingual gland, buccal mucosa and floor of the mouth.[2] Adenoid cystic carcinomas are clinically innocuous lesions usually characterized by small size and slow growth.[3] However, they are generally associated with extensive subclinical invasion and distant metastasis.[3] Pain is an important symptom of the condition due to its propensity for perineural spread.[2] Thus, ACCs have a long clinical course and questionable prognosis[2,3] with minor salivary gland ACCs having a worse prognosis than those of the major salivary glands.[2] We describe the features of adenoid cystic carcinoma in the buccal mucosa along with a review of the literature.

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CASE REPORT

A 45-year-old female reported to the department with complaint of a painful swelling in the left buccal mucosal region. She first noticed the swelling 3 months ago, which had gradually increased in size. It was associated with pain, which was mild and continuous in nature. On intraoral examination, there was an ill defined swelling in the left posterior buccal mucosa in the molar region [Figure 1]. Palpation revealed a tender, well-defined, freely movable swelling, 1 × 1 cm in size, which was soft to firm in consistency. The overlying mucosa was normal.

Panoramic radiography revealed no evidence of bone changes in the maxilla or mandible [Figure 2]. Ultrasonography of the region showed a hypoechoic mass with uniform internal structure and well defined borders. There were no areas of calcification and it

appeared unattached to the neighbouring structures [Figure 3]. These features were suggestive of a well defined, benign soft tissue mass. Lesion was surgically excised. Histopathological examination revealed loss of cellular architecture and cribriform pattern of tumor cells with many microcytes. Perineural invasion was present [Figure 4]. Based on the above features adenoid cystic carcinoma was diagnosed. Since the surgical margins were free of the disease, it was decided not to give any adjuvant therapy. She was followed up for 3 years with no signs of recurrence.

DISCUSSION

Adenoid cystic carcinoma is a rare epithelial tumor with an indolent but persistent growth pattern.[4] The World Health Organisation defines ACC as a "basaloid tumor consisting of epithelial and myoepithelial cells in various morphological configurations, including tubular, cribriform and solid patterns. It has a relentless clinical course and usually fatal outcome".[5]

ACC occurs predominantly in fourth to sixth decade of life with a female predilection of 3:2. In our case a 45-year-old female was affected.[2] Among salivary gland neoplasms, 9 to 23% occurs intraorally, of which 50% are malignant.[3]

Most articles in the literature describing the incidence of adenoid cystic carcinoma include both the major and minor salivary glands and no article so far has compiled the number of cases of ACC of the buccal mucosa alone. Our article is the first to present data gleaned from a total of 41 published articles. Only those articles which were specific regarding the intraoral site of involvement were included in our review. Articles which were unclear regarding the site of occurrence were excluded. After compilation of the cases, we found 2,280 cases of ACC in a total of 41 articles. Out of these cases 1,382 were reported in intraoral sites and 178 were specifically reported

Figure 1: Clinical Intraoral picture showing swelling in the left posterior buccal mucosa (black arrow)

Figure 3: Ultrasonographic image showing a well defined hypoechoic mass with uniform internal structure (lesion extent marked with X)

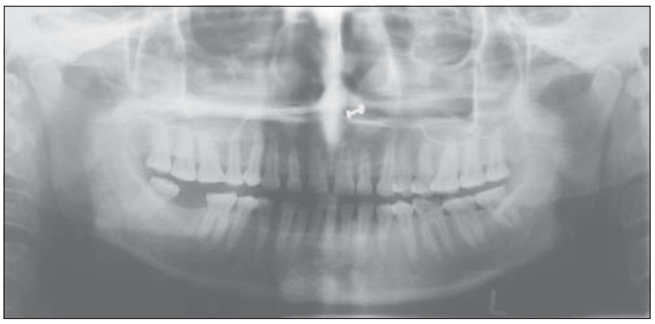

Figure 2: Orthopantomograph showing no bony changes

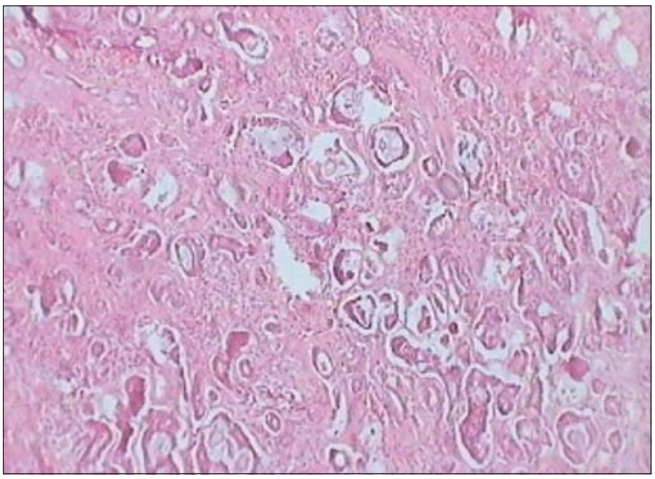

Figure 4: Photomicrograph ×10 showing cribriform pattern of tumor cells

in the buccal mucosa[2-4,6-43] [Table 1]. Based on the above findings, we concluded that among intraoral minor salivary gland ACCs, 12.9% occurred in the buccal mucosa. The overall percentage of buccal mucosal ACCs was 7.8%. According to the articles reviewed, 60.6% of all adenoid cystic carcinomas occur in an intraoral site.

The clinical presentation of ACC involves a slow growing, firm, unilobular mass.[42] Pain is usually a common and important associated symptom, occasionally occurring before clinical evidence of the disease.[42] Pain is often continuous and dull aching in nature. This case presented as a slow growing painful swelling. Local lymph node involvement is rare. Hematogenous spread occurs late in the course of the disease.[2,44] Distant metastasis is commonly to the lung, bone and soft tissues.[2]

Histologically, adenoid cystic carcinomas are of 3 types- cribriform, tubular and solid.[3,5] The tubular variant has the best prognosis and the solid variant the worst. The cribriform variant has intermediate differentiation and prognosis.[5] ACCs are graded according to the histological pattern into grade I, grade II and grade III with Grade I being a combination of cribriform and tubular, Grade II a mixture of cribriform, tubular and solid patterns and Grade III having only solid pattern.[3] The present reported case was of the cribriform variant and was classified as Grade I. Tendency to show perineural invasion is a highly characteristic feature of ACC. Common clinical feature of pain in these patients may be due to perineural invasion. However, it is not a pathognomonic feature of the disease.[42] Perineural invasion occurs through spread along the perineural spaces or within the nerve itself.[5] According to WHO "the influence of perineural invasion on survival has been contradictory".[5] It was stated to have no prognostic significance in some studies[5] whereas some authors mention that it is a negative survival predictor because of greater tendency for distant metastasis.

Various treatment modalities that have been proposed in ACC which include surgery, radiotherapy, chemotherapy and combined therapy. Surgical excision with wide margins is the treatment of choice.[42] We have followed a similar treatment protocol. Many factors influence the prognosis in cases of adenoid cystic carcinoma. These include tumor stage, positive surgical margins, site of primary, perineural invasion, solid histological type and presence of cervical lymph node metastasis at the time of diagnosis.[3,42,45] ACCs typically have a prolonged clinical course with distant metastasis occurring late in the disease despite adequate locoregional control.[2,42] One study discovered that the median time between diagnosis of the primary lesion and detection of distant metastasis was 60 months with a range of 18-120 months.[4]

Table 1: Reported cases of adenoid cystic carcinoma of the buccal mucosa

Author/Year	Total cases	Intraoral minor salivary gland	Buccal mucosa
Abaza *et al.* 1966[6]	3	3	0
Ampil *et al.* 1987[7]	31	16	2
Hosokawa *et al.* 1992[8]	41	20	2
Shick *et al.* 1995[9]	12	3	0
Ishikawa *et al.* 1997[10]	1	1	0
Huang *et al.* 1997[11]	91	38	3
Torre *et al.* 1997[12]	1	1	0
Umeda *et al.* 1999[13]	30	30	4
Dori *et al.* 2000[14]	27	14	1
Umeda *et al.* 2000[15]	17	11	4
Huber *et al.* 2001[16]	75	23	4
Kiyoshima *et al.* 2001[17]	17	13	3
Chummun *et al.* 2001[18]	45	5	0
Okamura *et al.* 2002[19]	18	13	3
Kulczynski *et al.* 2003[20]	135	34	9
Enamorado *et al.* 2004[21]	46	11	3
Goodwin *et al.* 2004[22]	1	1	1
Wang *et al.* 2005[23]	4	4	1
Freier *et al.* 2005[24]	27	7	1
Rapidis *et al.* 2005[4]	23	13	0
Luo *et al.* 2006[25]	20	6	0
Giannini *et al.* 2006[2]	1	1	1
Da Cruz Perez *et al.* 2006[26]	129	54	5
Wang *et al.* 2007[27]	143	143	17
Hirota *et al.* 2007[28]	4	4	0
Ferazzo *et al.* 2007[29]	14	13	6
Buchner *et al.* 2007[30]	24	24	1
Greer *et al.* 2007[31]	39	31	4
De Noronha Santos Netto *et al.* 2008[32]	3	3	1
Subhashraj *et al.* 2008[33]	66	25	7
Mucke *et al.* 2009[34]	33	33	8
Tilakratne *et al.* 2009[35]	96	81	8
Ahmed *et al.* 2010[36]	35	26	0
Tian *et al.* 2010[37]	681	403	36
Isa Kara M *et al.* 2010[38]	11	5	0
Li *et al.* 2010[39]	63	27	7
Xinjie *et al.* 2010[40]	72	41	4
Martinez-Rodriguez *et al.* 2011[41]	193	193	31
Gondivkar *et al.* 2011[42]	1	1	0
Mahajan *et al.* 2011[3]	1	1	0
Matsuzaki *et al.* 2011[43]	6	6	1
Total	2280	1382	178

Unlike other malignancies, they usually do not lead to death in the short term[46] but have low long term survival rates.

CONCLUSION

Adenoid cystic carcinomas are seemingly innocuous lesions, which show slow growth but due to their propensity for perineural spread and distant metastasis, require prolonged follow-up.

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