Adenosquamous Carcinoma of Buccal Mucosa
— A Case Report

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Abstract

This paper reports a rare case of adenosquamous carcinoma that involved the left buccal mucosa in a 47-year-old man, with rapidly tumor recurrence in two month after an initial surgery. Adenosquamous carcinoma is a very rare malignant tumor with poor prognosis that characterized histopathologically by the simultaneous presence of distinct areas of adenocarcinoma and squamous cell carcinoma. Only 59 cases of adenosquamous carcinoma within the head and neck region have been documented in the English literature since 1968. Although most of the previous report showed very poor prognosis with these tumor, fortunately, after two major radical operations and without post-operative radiotherapy, our patient still maintains a good condition with no further evidence of tumor recurrence and metastasis for more than five years after surgery.

Key words: adenosquamous carcinoma, buccal mucosa, oral cavity.

INTRODUCTION

Adenosquamous carcinoma is a very rare, aggressive, malignant tumor in the oral cavity that has distinct squamous and glandular components histologically. Less than sixty cases have previously been reported in the oral cavity, but all have shown an aggressive course with 60% of patients dying of such a disease. The histogenesis of this malignant tumor, a high-grade variant of malignant epithelial neoplasm, has long been debated. We are now reporting a case of adenosquamous carcinoma of the buccal mucosa, which presented as an aggressive tumor initially, but for which after two radical operations have resulted in a good and stable condition after five years.
CASE PRESENTATION

A 47 year–old man visited our dental clinic on Nov. 2002 due to a painful large exophytic tumor mass over his left cheek and facial swelling for several months. The clinical examination revealed an irregular diffuse reddish exophytic tumor mass over his left buccal mucosa to posterior lower gingiva about 5 × 6 cm in size. An extra–oral examination showed a moderate swelling over his left cheek with an obvious facial asymmetry (Fig. 1). Under the impression of malignancy, an incisional biopsy was performed on the same day. However, the pathological report showed only a diagnosis of pyogenic granuloma. A CT–scan was then arranged because of highly suspicious of malignancy from the clinical standpoint. The image from the CT–scan showed a cystic lesion about 2.7 × 4.6 cm in size, containing an enhanced solid part in the left buccal space and adjacent masticator space (Fig. 2). Based on the pertinent clinical information, the differential diagnosis of the malignancy included buccal carcinoma and inflammatory pseudo–tumor as well as a combined carcinoma and inflammatory process was reported. Under the impression of malignancy, the patient was arranged admission for surgical intervention. Due to the initial biopsy report showed only a pyogenic granuloma, a piece of tissue within the tumor mass was taken out for a frozen section first before the operation. The frozen section showed squamous cell carcinoma this time. The operation was then started with an en bloc tumor excision with 1cm safe margin from the facial skin through and through to buccal mucosa and gingiva with marginal mandibulectomy as well. The post excision defect over the left face and cheek was reconstructed with a double paddle, free peroneal flap from the left leg. The whole procedure was smooth and uneventful. The final pathology report was adenosquamous carcinoma, moderately differentiated, with skin and left mandible invasion. After being hospitalized for twenty days, the patient was discharged in a stable condition. Unfortunately, after two months post–operative follow–up, a fistula with discharge from the previous repaired flap margin over the left face was noted. Mild swelling over the left side mouth floor and the posterior cheek as well, but the intra–oral mucosa was intact. Under the impression of R/O tumor recurrence, some soft tissue was removed from the fistula and sent for pathology again. The pathological report confirmed a recurrent adenosquamous carcinoma. The patient was arranged another admission for a further evaluation with CT–scan and operation. This time the CT report showed a soft tumor mass about 4.2 × 5.6 cm in size over his left buccal region and left masticator space and this mass was more in favor of a tumor recurrence. No obvious lymphadenopathy over bilateral neck was found. (Fig. 3). A further wide excision with supra–omohyoid neck dissection and hemi–mandibulectomy was performed. Another free fibula composite flap with a double skin paddle and 8 cm of fibula bone was taken from right leg for wound defect reconstruction. After the second operation, the patient stayed in the hospital for 21 days and was discharged in good condition. Further post–operative radiotherapy was suggested, but the patient declined. After that, the patient has remained well with no evidence of tumor recurrence or distant metastasis for more than 5 years follow up. (Fig. 4).
Pathological findings

The resected specimen from the first operation, a wide excision of the left buccal mucosa and a partial resection of left mandible, showed a papillary lesion 4.5 × 4.0 × 4.0 cm on the mucosa, which extended to the adjacent soft tissue and also the mandible. The diagnosis is adenosquamous carcinoma, composed of both malignant squamous and glandular components (Figs. 5,6,7) with the presence of squamous carcinoma in situ (Fig. 8). The surrounding soft tissue and the mandible are also involved. The second operation tissue specimen, a wide excision and neck dissection, showed a gray white firm mass lesion 5.5 × 5.0 × 4.5 cm on the mucosa, which extended to the underlying mandibular bone. The pathological finding also showed adenosquamous carcinoma, with bone invasion but no lymph node metastasis.

DISCUSSION

Adenosquamous carcinoma is a rare, controversial malignant tumor because of the unknown histogenetic origin. Until 2004, only 59 cases of adenosquamous carcinoma within the head and neck region had been reported in the English literature since 1968. Various authors have postulated different theories regarding the histogenetic origin of adenosquamous carcinoma in the oral cavity. Some authors have suggested that the adenosquamous carcinoma is a malignant salivary gland tumor. However, the 1991 WHO classification of salivary gland tumors did not include this malignancy among the tumors of salivary glands. Some authors considered that the tumor might be originating from the reserve or basal cells of the covering squamous epithelium which underwent divergent differentiation. An experimental model of adenosquamous carcinoma has recently been developed, suggesting that this tumor does not originate from salivary or seromucous glands and providing support for its origin from the reserve cells of the squamous epithelium.

Since 1997, the World Health Organization has defined adenosquamous carcinoma as a malignant tumor characterized by the simultaneous presence of both adenocarcinoma and squamous carcinoma, with the two components occurring in close proximity, but generally distinct. The most common site of origin within the head and neck region has been from the larynx, accounting for 44.8% of all cases. The oral cavity has been the second most common site of origin, of which the floor of mouth and the tongue are the dominant sites. Nevertheless, so far, there has been no case reported arising from the buccal mucosa, and the case we report here is the first case described in this region. The literature review by Keelawat et al. indicated that the clinical course of adenosquamous carcinoma is characterized by local recurrence, cervical lymph node metastasis, distant metastasis, and a poor long term prognosis. 46.7% of patients had local recurrences, 64.7% developed cervical lymph node metastasis, and 23.1% experienced distant metastasis. The overall 5-year survival rate was 13%, and 42.9% of patients died at a mean follow-up period of 24.7 months. According to previous cases reported, it implies that adenosquamous carcinoma is an aggressive tumor, which always spreads to the cervical lymph nodes and shows distant metastasis after treatment.

Although we encountered initial difficulty
Fig. 1. Intra-oral and extra-oral view of the patient.

Fig. 2. Initial CT-scan images showing a cystic lesion with containing enhanced solid part noted in left buccal space and adjacent masticator space.
Fig. 3. 2nd CT-scan images showing large recurrent tumor mass over previous operation site.

Fig. 4. Five years post-operative extra-oral and intra-oral view of the patient.
Fig. 5. Adenosquamous carcinoma of buccal mucosa with extensive keratinization and necrosis. H&E, x 40.

Fig. 6. Adenosquamous cell carcinoma with distinct malignant squamous (left) and glandular (right) components.
Fig. 7. Adenosquamous cell carcinoma with mucin pool.

Fig. 8. Adenosquamous carcinoma of buccal mucosa. Presence of both invasion (broad arrow) and in situ (narrow arrow) features.
in diagnosis from the tissue biopsy and also from the frozen section during operation, the definitive diagnosis was made after the whole surgical specimen sent to the pathology department. The difficulty of the diagnosis of our case can be explained by the significant but small proportion of the adenocarcinoma component, which was missed from the biopsy and frozen section. The main differential diagnosis of adenosquamous carcinoma is mucoepidermoid carcinoma. In the past, some authors did not distinguish between adenosquamous and high-grade mucoepidermoid carcinoma. There are five differential criteria:

a) Presence of squamous cell carcinoma in situ mostly in the surface epithelium and occasionally in the ducts of the seromucous glands, a feature typical of adenosquamous carcinoma and absent in mucoepidermoid carcinoma.

b) Prominent separated foci of squamous cell carcinoma and adenocarcinoma. In mucoepidermoid carcinoma both patterns are closely intermingled.

c) Foci of squamous epithelium with marked keratinization in adenosquamous carcinoma. Keratinization is not seen in the epidermoid component of mucoepidermoid carcinoma.

d) Intermediate cells, which are typical of mucoepidermoid carcinoma, are absent in adenosquamous carcinoma by light microscopy.

e) Severe nuclear pleomorphism and atypicality in adenosquamous carcinoma. Our case fits all the criteria described above. High-grade mucoepidermoid carcinoma is less atypical and less aggressive than adenosquamous carcinoma. It also shows a better survival than adenosquamous carcinoma. Although we have performed a radical surgery initially, the tumor rapidly recurred and grew faster than the first time. This implied the aggressive behavior of this kind of tumor as other authors suggested. However, the case we have reported here did not have any regional lymph node metastasis, which seems inconsistent with the clinical course described by Keelawat. This may suggested that a case without lymph node involvement may have a better prognosis than a case with lymph node metastasis. Moreover, even without post-operative radiotherapy due to patient refused to do so, our patient still remained tumor-free and survived more than 5 years after the second operation.

In summary, adenosquamous carcinoma is a rare but aggressive tumor in the oral cavity. Most of the previously reported cases are associated with a high rate of locoregional recurrence and death from disease after surgical treatment in a short period. However, our case arising from buccal mucosa in a 47-year-old man is unusual for the location, but has nonetheless displayed a rapid recurrence as the others, but fairly good prognosis after two radical operations for more than five years.

REFERENCES


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摘 要

本文報告一罕見腺樣鱗狀細胞癌發生於47歲男性病患左頰黏膜之病例，腺樣鱗狀細胞癌極為少見且預後不佳，同時出現腺體癌及鱗狀細胞癌之病理特徵。自1968年以來只有59例發生於頭頸部之病例報告於英文期刊上，雖然過去的報告顯示此癌症之預後極差，但本病例經兩次徹底手術後、經五年追蹤並無發生局部復發及遠端轉移的情形，因此特就此癌症之臨床特徵及病理診斷依據作一介紹及討論。

關鍵詞：腺樣鱗狀細胞癌，頰黏膜，口腔。