ADENOSQUAMOUS CARCINOMA OF HARD PALATE: A CASE REPORT

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ABSTRACT

Adenosquamous carcinoma of the head and neck (ASC) is a rare and aggressive variant of squamous cell carcinoma, a locally aggressive malignancy characterized by the presence of two distinct components: a squamous cell carcinoma and an adenocarcinoma. The purpose of this article was to report an additional rare case of adenosquamous carcinoma affecting the maxilla.

Keywords: Adenosquamous carcinoma, oral cavity, hard palate, maxilla.

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INTRODUCTION

Adenosquamous carcinoma (ADSC) is a rare entity of head and neck squamous cell carcinoma (SCC) which is characterized by mixed differentiation, with both SCC and true adenocarcinoma, as stated by the World Health Organization[1]. It has been described in various body sites, including the uterine cervix, lung, and pancreas. The extent of ADSC of the head and neck as a distinct entity was somewhat argumentative for many years as some investigators considered it to be a high grade mucoepidermoid carcinoma[2]. It was first defined in the head and neck byGerughty et al. in 1968 where itshowed extreme aggressiveness and highly malignantnature, with 80% of the patients having proven metastases[3]. The distinctly worse prognosis of ADSC as compared to mucoepidermoid carcinoma (MEC) necessitated the separation of these entities.

CASE REPORT

A 73-year-old male had been referred to our outpatient department with a chief complaint of an ulcer on hard palate noticed about two months before. The patient had a habit of smoking and drinking alcohol for a period 50 years with unremarkable medical history, had an ill-fitting maxillary complete denture. On examination a well-defined ulcer of size 1.5 cm india meter with everted margins extending to soft palate (fig 1) was noted. The lesion is erythematous and oval in shape. On palpation it is painless, indurated and no lymph nodes noted. Based on these findings, a provisional diagnosis of carcinoma of palate was given. Incisional biopsy of the lesion was done under local anesthesia using 2% lignocaine and 1: 80,000 adrenaline.

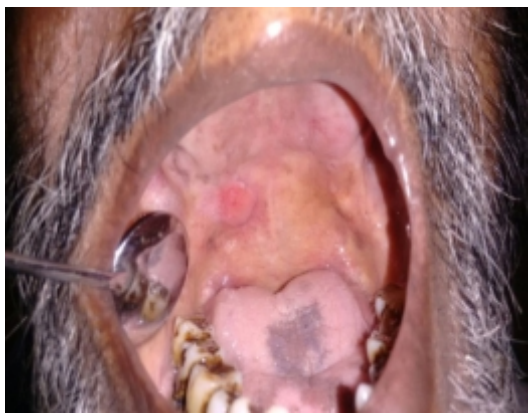


Figure 1 : Lesion in palate

Histopathological examination of incisional biopsy specimen showed dysplasia and marked hyperplasia, from which apparently an epithelial neoplasia had developed (Fig. 2A). This neoplasia was characterized by proliferation of atypical keratinocytes, forming islands of neoplastic cells infiltrating the connective tissue in the lamina propria, consistent with well-differentiated squamous cell carcinoma (Fig. 2B). Often, in the center of these islands, keratin pearls were observed, occasionally with superposed dystrophic calcification. Also in the central portions, some islands presented acantholysis producing pseudoluminae that were empty or contained cellular debris. Individually, neoplastic cells showed moderate pleomorphism, mitotic figures, multiple evident nucleoli, and marked dyskeratosis.

In deeper portions of the lesion, a distinct glandular neoplasia could be observed (Fig. 2C), showing tubular structures limited by two or more layers of epithelial cells, frequently containing eosinophilic pale material (Fig. 2D). The stroma consisted of dense connective tissue with capillary blood vessels, showing diffuse mononuclear inflammatory infiltrate and, in some areas, myxoid features. The lesion margins were poorly defined, with highly infiltrative aspect in the deeper front of invasion. Peripheral portions of the specimen showed minor salivary glands apparently not affected by the tumor.

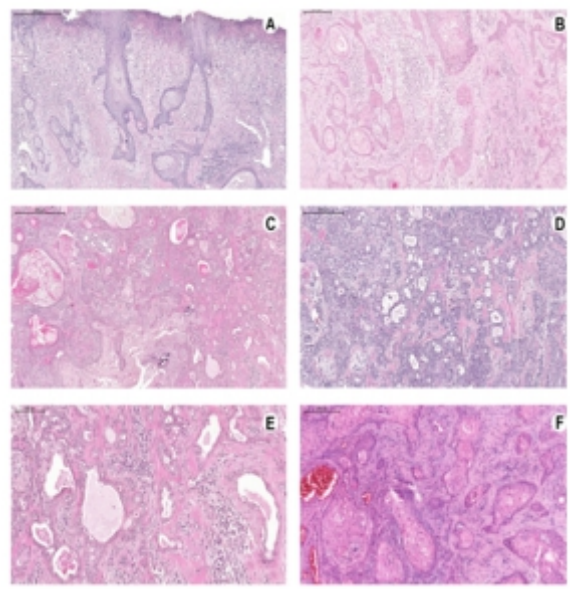
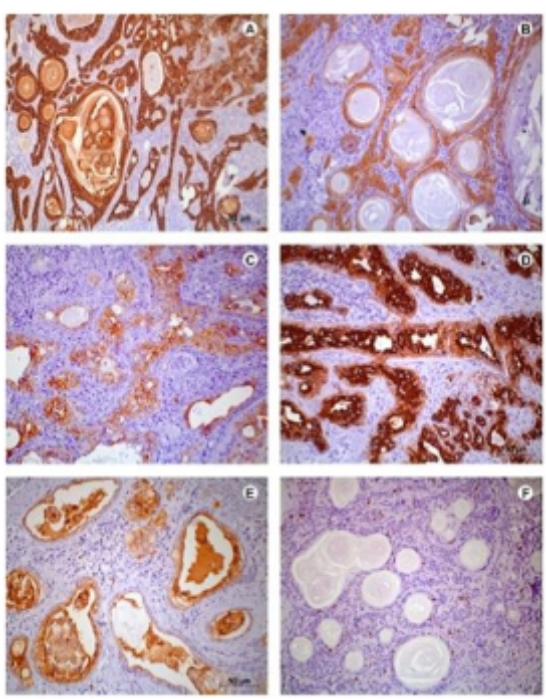


Figure 2: Histopathologic evaluation showed dysplastic stratified squamous epithelium proliferating into connective tissue in a nest, island and duct like

The histological hypothesis included squamous cell carcinoma, mucoepidermoid carcinoma, and pleomorphic adenoma. Throughout the diagnostic process, the hypothesis of acantholytic squamous cell carcinoma and of adenosquamous carcinoma were also included.

Histochemical staining with PAS, PAS-diastase, and mucicarmine only evidenced the content of the pseudoluminal structures. Immunohistochemical and histological features rendered the diagnosis of adenosquamouscarcinoma. All immuno histochemistry reactions were done using humid heat for target retrieval and streptavidin-biotin-peroxidase system for detection (fig 3). Positive and negative controls for every antibody were also included.

Figure 3: Immunohistochemistry showed intraductalsialomucin positivity with mucicarmine special stain.



DISCUSSION

This case conferred very distinct clinical and microscopic features, hence several diagnostic hypothesis were arrived both at clinical and histological examination. Our first clinical hypothesis was squamous cell carcinoma as there are no other cases of adenosquamous carcinoma reported in hard palate with clinical findings similar to the one reported in our department. Even though in our case the evidence of malignancy has not been found clinically, the hypothesis of mucoepidermoid carcinoma could be raised, since this entity occurs frequently in hard palate and in a wide range of ages. Mucoepidermoid carcinoma, other than the classical submucosal bluish growth with fixed borders may present with several clinical appearances such as benign lesions. The benign salivary gland tumour with highest incidence in hard palate is certainly pleomorphic adenoma affecting young to middle-aged adults, presenting as a painless, slow growing mass. These are frequently located laterally in posterior hard palate and the mass usually presents regular surface [4]. In the present case, the mass was lobulated and located centrally in hard palate with an ulcerative component in the centre.

On histopathological assessment, the initial diagnosis of the lesion was a well-differentiated squamous cell carcinoma, since almost all features under light microscope were consistent with that. This diagnosis was not concluded due to the deeper part of the specimen showing adenoid neoplastic components which were obviously not part of the surrounding normal minor salivary glands.

Mokhtari et al.[5] reported a similar situation, in which the initial histopathological diagnosis was squamous cell carcinoma and on further evaluation the adenocarcinomatous component was noticed in metastatic cervical lymph nodes. In the same report the diagnosis was then changed to adenosquamous carcinoma when discrete neoplastic gland-like structures were identified using specific histochemical tests. Likewise, Fonseca et al. described a lesion initially diagnosed as epithelial dysplasia with focal microinvasive well-differentiated squamous cell carcinoma and after 12 months of follow-up a local recurrence was noted which presented with clinical appearance different from the initial lesion and it showed features consistent with adenosquamous carcinoma [6].

The mostcommon site of occurrence for adenosquamous carcinoma in the head and neck region is larynx with 48.4% followed by the oral cavity at 30%[7].The most common locations in the oral cavity are the floor of the mouth, tongue, alveolus, palate and upper lip [8]with 6:1 male to female ratio. Differential diagnosis for adenosquamous carcinoma comprises ofmucoepidermoid carcinoma, acantholytic squamous cell carcinoma, basaloid squamous cell carcinoma, conventional squamous cell carcinoma and necrotizing sialometaplasia.

The main histological differential diagnosis associated with this is mucoepidermoid carcinoma. In mucoepidermoid carcinoma gland-like structures and squamous cells tend to be intermingled while in adenosquamous carcinoma these two components are often separate and appear as two distinct areas of the tumor[7]. Pleomorphic adenoma was also considered in the histological differential diagnosis, due to areas presenting tubular structures limited by two or more layers of cells, often presenting a mucinous content. However, this hypothesis was discarded with the observation of clear and exuberant cellular pleomorphism, mainly seen in the squamous component of the analyzed specimen.

In this casethe diagnosis of squamous cell carcinoma seemed to be appropriate other than the two minor features observed under light microscope, which includes: acantholysis in a small number of neoplastic squamous islands forming pseudoluminae, containing or not cellular debris, and the adenocarcinomatous component, which apparently is not a part of the normal minor salivary glands of the hard palate. Hence, two other histological hypotheses were then considered: acantholytic squamous cell carcinoma and adenosquamous carcinoma, respectively. Acantholytic squamous cell carcinoma is another uncommon variant of squamous cell carcinoma, characterized by acantholysis of neoplastic squamous cells, creating pseudoluminae and a false appearance of glandular differentiation[8]. In the present case a few areas were seen with these pseudoluminae.

Likewise, in immunohistochemical analysis, the present case was immunopositive for carcinoembrionic antigen, excluding acantholytic squamous cell carcinoma and emphasizing the final diagnosis of adenosquamous carcinoma[8]. In the present case immunohistochemical findings were crucial to reach the final diagnosis of adenosquamous carcinoma. AE1/AE3 stained positive in all neoplastic cells and in the surface epithelium, indicating the epithelial origin of the lesion (fig 3A). However when considering different molecular weight keratins separately, it became obvious the dual histomorphogenesis of the neoplastic cells. That was confirmed by the positive staining for CK5 (high weight) in the squamous component (fig 3B) and the positive staining for CK8/18 (low weight) in the adenocarcinomatouscomponent (fig 3C). Staining to CK7 (low weight) was strongly positive in the cells in proximity with the luminae (Fig. 3D). Carcinoembrionic antigen is a marker largely used for the final diagnosis of adenosquamous carcinoma. The present case showed scarce positivity in the squamous component and intense positivity in the adenocarcinomatous component (Fig. 3E).The proliferation index based on Ki67 immunostaining was considered low (approximately 10%) for a malignant lesion of known aggressive behavior (Fig. 3F). Study conducted by Keelawat et al.[9] noted 47% of patients with local recurrence, 65% nodal metastasis and 23% distant metastasis and 42.9% died of their disease at a mean follow-up period of 24.7 months. Treatment strategy varies with chemotherapy, surgery and radiotherapy based on the site and stage of disease. After achieving a diagnostic conclusion our case was referred to Regional cancer centre for appropriate treatment and follow up.

CONCLUSION

Adenosquamous carcinoma represents a diagnostic challenge due to its diverse range of clinical presentations and histological features asitsadenocarcinomatous component may be, at times, difficult to identify. The recognition of the specific subtype of squamous cell carcinoma is of great significance.

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