



Adenosquamous Carcinoma of The Floor of The Mouth: A Case Report and Review of Literature

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Abstract

Adenosquamous carcinoma (ASC) is an aggressive variant of squamous cell carcinoma (SCC) with high infiltrative capacity. It is distinguished by the coexistence of both squamous cell carcinoma and adenocarcinoma areas. Adenosquamous carcinoma (ASC) is infrequently found in the upper aerodigestive tract, and it is especially rare in the oral cavity. The treatment of choice is surgery combined with neck dissection. To our knowledge, there are very few documented cases of ASC in the floor of the mouth. Hereby, we present an additional case involving a 68-year-old female patient with ASC located in the floor of the mouth.

Keywords: Adenosquamous carcinoma; floor of the mouth; squamous cell carcinoma

Introduction:

Adenosquamous carcinoma (ASC) is a rare variant of squamous cell carcinoma (SCC) notable for its high infiltrative ability and tendency to metastasize to lymph nodes. According to the WHO 1995 classification of tumors of the upper respiratory tract and ear, ASC of head and neck region was defined as "A malignant tumor with histological features of both adenocarcinoma and SCC." [1]. Adenosquamous carcinoma (ASC) is also considered as a controversial tumor due to its similarity to salivary gland mucoepidermoid carcinoma (MEC). However, Evans, in 1984, emphasized that ASC of the head and neck has a worse prognosis compared to high-grade MEC. Consequently, he proposed that ASC should be classified as a distinct neoplasm [2]. The most common sites for adenosquamous carcinoma in the oral cavity are the tongue, followed by the palate, tonsillar pillar areas, and floor of the mouth [3]. In this article, we report a rare case of adenosquamous carcinoma of the floor of the mouth in a 68-year-old female patient.

Case report:

A 68-year-old female patient with no notable medical history was referred to the Department of Oral and Maxillofacial Surgery of the University Hospital Center Hassan II with a 3 years history of a swelling in the the floor of the mouth. Extraoral examination revealed no facial asymmetry. Intraoral Examination revealed a 2.5x2 cm indurated mass in the floor of the mouth extending from the 42 tooth region to the 34 tooth region (**figure 1**); the lesion is erythematous and irregular in shape with lobular surface. There were no extension to the ventral surface of the tongue and no palpable cervical lymph node. Based on these findings, a provisional diagnosis of carcinoma of the floor of the mouth was given and incisional biopsy was performed.



Figure 1: Intra oral picture showing a mass in the floor of the mouth extending from the 42 tooth region to the 34 tooth region.

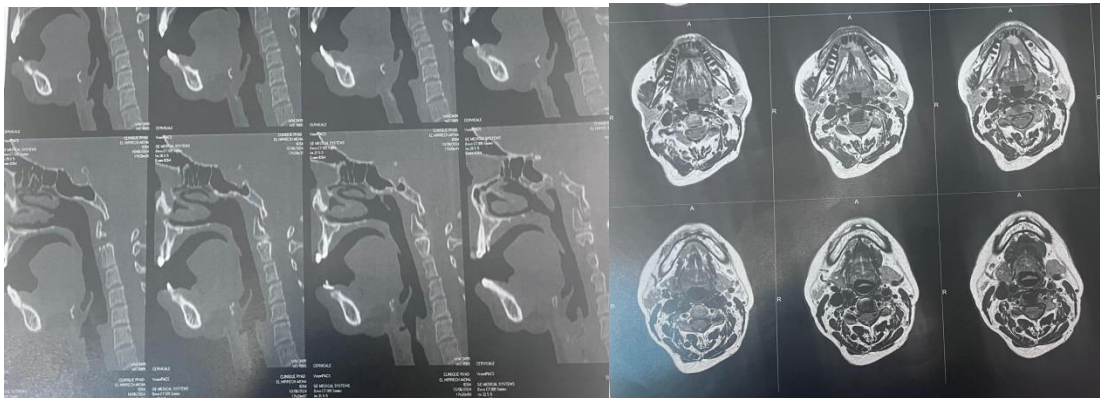


Figure 2: Sagittal CT scan showing a lytic lesion of the floor of the mouth at the lingual surface of the mandibular symphysis (a), axial MRI scan revealing a tumor of the floor of the mouth measuring about 2.5×2.0 cm, infiltrating the cancellous bone of the mandible.

Based on the Histopathological examination of the incisional biopsy, diagnosis of adenoid cystic carcinoma was established. Cervico-facial CT scan and facial MRI scan were performed revealing a tumor of the floor of the mouth measuring about 2.5×2.0 cm, infiltrating the cancellous bone of the mandible and exerting a mass effect on the sublingual glands and the termination of the Wharton's duct, but no cervical lymph node swelling

Subsequently, the patient underwent a non interruptive pelvimandibulectomy with reconstruction of the defect by a left naso labial flap and bilateral neck dissection (level I – IV). The excised specimen was sent for histopathological examination. The histopathological examination showed that the oral mucosa was

covered by dysplastic parakeratinized epithelium. The underlying connective tissue was infiltrated by nests and strands of malignant epithelium, which exhibited central keratinization. Additionally, the salivary ducts involved displayed significant dysplasia and adenocarcinomatous changes with mucus production (**figure 3 a;b**). The tumor consisted of two distinct components: a superficial squamous component and a deeper glandular component; the features were consistent with adenosquamous carcinoma.

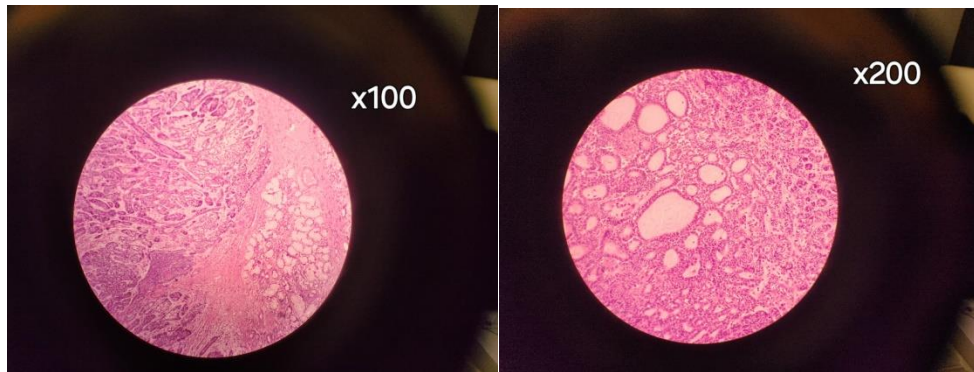


Figure 3: Histological image showing squamous and adenocarcinomatous regions with obvious mucus-producing cells (a×100, b×200).

The resection specimen confirmed the diagnosis of adenosquamous carcinoma, and the surgical margins were clear of any tumor. Following the surgery, the patient received adjunct radiotherapy (figure 4, 5). As of the time of this report, the patient has been under follow-up for two years, and there have been no signs of a recurrence.



Figure 4: Extra oral picture showing the scars from the donor site of the left naso labial flap (a) and the left neck dissection (b).



Figure 5: Intra oral picture showing the reconstruction of the defect related to the pelvimandibulectomy by a left naso labial flap.

Discussion:

Adenosquamous carcinoma is an uncommon and aggressive malignant epithelial neoplasm of the oral cavity. According to the

WHO, adenosquamous carcinoma is a type of malignant tumor that features both true adenocarcinoma and squamous carcinoma components. These components are typically found near each other but remain distinct from one another [4].

Similar to conventional squamous cell carcinoma, alcohol and tobacco abuse may also be significant risk factors for adenosquamous carcinoma (ASC). There are various theories about the histogenesis of adenosquamous carcinoma in the oral cavity. Some authors propose that this type of carcinoma originates from salivary gland cells [5], other authors suggest that adenosquamous carcinoma may originate from reversed or basal cells of the squamous epithelium, which then undergo various stages of differentiation [6]. Recent experimental model of adenosquamous carcinoma suggest that the tumor does not originate from salivary or seromucous glands. Instead, this models support the theory that it originates from the reserve cells of the squamous epithelium [7]. Recent research has also demonstrated that human papillomavirus (HPV) is an etiological agent in head and neck carcinomas, particularly in the oropharynx and nasal cavity.

In the head and neck region, adenosquamous carcinoma (ASC) is most commonly found in the tongue, palate, floor of the mouth, and larynx with the highest incidence occurring in the fifth decade [8]. A common complaint among patients presenting to clinicians is pain, which is often due to the tendency of the neoplasm to spread through perineural invasion. Clinically, it can present as a keratotic ulcer or as nodular, exophytic, indurated, or irregularly shaped masses.

According to the WHO definition, adenosquamous carcinoma (ASC) displays dual histomorphology, featuring both squamous and true adenocarcinoma components. [9]. The squamous cell component typically predominates and is found in the superficial areas, originating from the surface epithelium. The mucosal epithelium shows severe dysplasia or carcinoma in situ, the presence of keratinization foci of varying extent was also observed [2]. The adenocarcinoma component is typically located in deeper areas and is composed of ductular structures containing varying numbers of mucous cells [9].

Immunohistochemically, adenosquamous carcinoma is positive for CEA in 92% of cases, CK7 in 75%, and CAM 5.2 in 58%. In contrast, squamous carcinoma is typically negative or only focally reactive for these markers. Both adenosquamous carcinoma and squamous carcinoma are positive for high molecular weight cytokeratin 34BE12. The differential diagnosis for adenosquamous carcinoma includes mucoepidermoid carcinoma, acantholytic squamous cell carcinoma, basaloid squamous cell carcinoma, conventional squamous cell carcinoma, and necrotizing sialometaplasia.

Adenosquamous carcinoma (ASC) is an aggressive tumor that can spread to cervical lymph nodes and is prone to locoregional and distant recurrence after treatment. [10]. Treatment typically involves surgical resection of the tumor and regional lymph nodes, which may be supplemented with radiotherapy [11, 12, 13]. Adenosquamous carcinoma is associated with a high metastatic rate of approximately 80% and a low 5-year survival rate of around 20–25% [14].

Conclusion:

Adenosquamous carcinoma (ASC) is a rare and aggressive

malignant neoplasm with a poorer prognosis compared to conventional squamous cell carcinoma (SCC). It is also associated with a high rate of lymph node metastasis.

Conflict of Interest:

The authors declare no conflicts of interest.

Authors' Contributions:

All authors participated in the patient's care and in the writing of the article. All authors have read and approved the final version of the manuscript.

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