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Up-to Date Review And Case Report

Adenosquamous carcinoma, a rare and unknown tumor

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Abstract – Introduction: Adenosquamous carcinoma (ASC) of the head and neck is a rare malignant tumor, with one hundred cases diagnosed so far in the literature. **Observation:** A 66 years old female patient had a ASC of the right posterior floor of the mouth with a classification T1N0M0. Treatment consisted of surgical management and active surveillance. **Discussion:** Through this case report, we present this type of tumor and we discuss the main differential diagnosis from a clinical and histological point of view. **Conclusion:** This entity was described for the first time in 1968 by Gerughty. The CAS is defined by the World Health Organization as “a tumor of the upper respiratory tract, with two distinct squamous and glandular components.” The CAS is considered as an aggressive tumor with a redoubtable prognosis should not be ignored.

Introduction

Adenosquamous carcinoma (ASC) is a rare and malignant tumor of the head and neck region. Approximately 100 cases have been reported in the literature. This entity was first described in 1968 by Gerughty [1]. ASC is defined by the World Health Organization as “a malignant tumor of the upper aerodigestive tract, which has two distinct squamous and glandular contingents.” The squamous part is usually predominant. It can manifest as a well differentiated or a poorly differentiated tumor, and may occur as a *carcinoma in situ* or an invasive tumor. The glandular component is most often found in the deeper layer of the tumor. ASC is an aggressive tumor with a poor prognosis with frequent local and locoregional metastases despite appropriate treatment [2].

Clinical case

A 66-year-old woman with no relevant medical or surgical history consulted us with complaints of persistent pain under the tongue. She was an active smoker at 40 pack years without any history of alcoholism. The pain had gradually worsened

over the course of 4 years. Intraoral examination revealed a wide erythematous region of the right posterior floor, rising to the lingual slope of the distally edentulous mandibular crest of 44. There was an ulceration in the area between the floor of the mouth (FOM) and the ventral surface of the tongue. The ulceration was localized on the erythematous area, surrounded by keratotic areas that were whitish, inhomogeneous, and somewhat thick (Fig. 1). Although the lesion was tender, it did not bleed on contact and was not indurated. The lingual function examination was normal, and no swallowing or phonation disturbances were noted. The extraoral exam was normal and no lymphadenopathy or swellings were noted.

Due to the inhomogeneous aspect of the lesion, a biopsy was performed. The histopathological analysis of the biopsy made the diagnosis of ASC (Fig. 2). The entire biopsy sample consisted of tumor material and showed the presence of a well-differentiated epidermoid component; one part was a *carcinoma in situ* and the other part was invasive, with a keratinized area and some keratin pearls. The glandular contingent consisted of four glands. There was also a large inflammatory infiltrate and the existence of many mitotic figures. Staining with periodic acid/Schiff (PAS) and alcian blue confirmed the secretory nature of these glands.

The patient was therefore referred to the head and neck cancer surgery department. In the preoperative assessment at the department, she underwent a pan-endoscopy of the upper

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aerodigestive tract, a cervicofacial and thoracic CT scans, and a cervicofacial MRI to accurately characterize the lesion (8 mm transverse diameter by 10 mm vertical diameter). The tumor was adjacent to the mandible without bone infiltration and there was no sign of lymph node or distant metastases. The lesion was classified as a cT1N0M0. Surgical intervention was decided upon at a multidisciplinary consultation meeting. A right posterior uninterrupted pelvi-mandibulectomy of 3.5 × 5.5 × 1.5 cm was performed associated with a complete ipsilateral cervical dissection, followed by posterior-based buccinator myomucosal flap reconstruction.

The histopathological analysis of the specimen confirmed the adenosquamous nature of the lesion with an ulcerated superficial, infiltrating tumor with a double component of massive, trabecular and glandular, cribriform architecture. It presented with a contingent of moderately differentiated squamous cell carcinoma with slight keratinization and masses of eosinophilic cells, which were surrounded, at some instances, by keratinization foci. Alcian blue staining revealed a glandular mucosecretory component in the deeper layers of the infiltrating tumor. The tumor had a 14 mm diameter at its major axis and showed capsular invasion of 1 mm. The stromal reaction was fibroinflammatory. There were many atypical mitotic figures (Fig. 3). Immunohistochemical analysis showed CK5/6 and CK7 positivity for the glandular component, and CK5/6 positivity and CK7 negativity for the epithelial component (Figs. 4 and 5). A test for human papillomavirus (HPV) was negative. The histopathological analysis classified the tumor as a pT1N0M0 R0 lesion (52 lymph nodes 103 analyzed) with a posterior section in dysplastic zone.

A complementary surgical procedure was therefore performed to widen the anterior and posterior margins. The analysis of the specimen did not find any tumor residuals, ruling out indications for adjuvant chemoradiotherapy treatment. The strict postoperative monitoring consisted of one clinical follow-up every 2 months over a 1-year period. Subsequently, the consultation frequency will be progressively spaced out. Regular computed tomography scans will be required as a means of radiological follow-up. This will occur every 6 months for 3 years because of the high risk of early relapse.

Discussion

ASC is considered an aggressive tumor with a guarded prognosis with up to 80% of patients having metastases [1]. Our patient had been reporting painful tumor episodes at more or less regular intervals for 4 years. Sheahan associates this symptomatology with frequent perineural invasions (found in 75% of the cases) [3], although they were not found in this case. The tumor was classified as cT1N0M0 under TNM staging, which was confirmed by histopathological analysis (pT1N0M0). The sex ratio (male: female) is usually between 9:1 [1] and 2:1 [4]. The age of occurrence in the present case corresponds to the median age of 60 years reported previously. [2].



Fig. 1. Macroscopic view of the lesion.

The oral cavity is the second most common location after the larynx [2]. The histopathological characteristics of this tumor and now well described. Unlike some previous report where the diagnosis was made only with the aid of a substantial immunohistochemical panel, [5,6] few antibodies were used for diagnosis in this case. The selective positivities CK5/6 and CK7 and the characteristic morphological features (superficial epithelial component, deep glandular) allowed for an early diagnosis. Thorough pathological examination, along with the known and compatible histopathological features, the histopathologist will seek to confirm the diagnosis by immunohistochemical examination. However, there is still doubt regarding whether the tumor had an epithelial or a salivary origin. Since its first description in 1968, when Gerughty [1] attributed a salivary origin to it, ASC was linked to the mucoepidermoid carcinoma (MEC). It was in 1984 that Evans [7] suggested separating these two entities in view of the poor prognosis of ASC, unlike that of MEC. The origin of this tumor is still being debated, and the evidence is pointing towards an epithelial origin. In 1991 Ellis [8] suggested an unprecedented double origin of ASC, *i.e.*, both salivary and epithelial. However, Napier [9] and Fonseca [10] have reported that origin is strictly epithelial.

Nevertheless, despite the precise histopathological description, this tumor may be incorrectly diagnosed. In effect, a superficial biopsy will not be able to reveal the glandular component. Moreover, its low frequency makes it possible to be confused with a MEC (Seethala states that 20% of high-grade MECs are in fact ASCs [11]), adenoid squamous cell carcinoma (ASCC) or basaloid squamous cell carcinoma (BSCC). In ASC, the squamous component is morphologically identical to MEC with the presence of keratinization foci. The glandular component is always formed from duct-like structures with or without mucus cells. These two components are always well separated. MEC is a salivary tumour with no macroscopic mucosal involvement. In the histopathological analysis of a high-grade MEC, a predominant presence of intermediate cells is observed without keratinization of any clear cells. In addition, glandular and squamous components show an overlap (Tab. I).

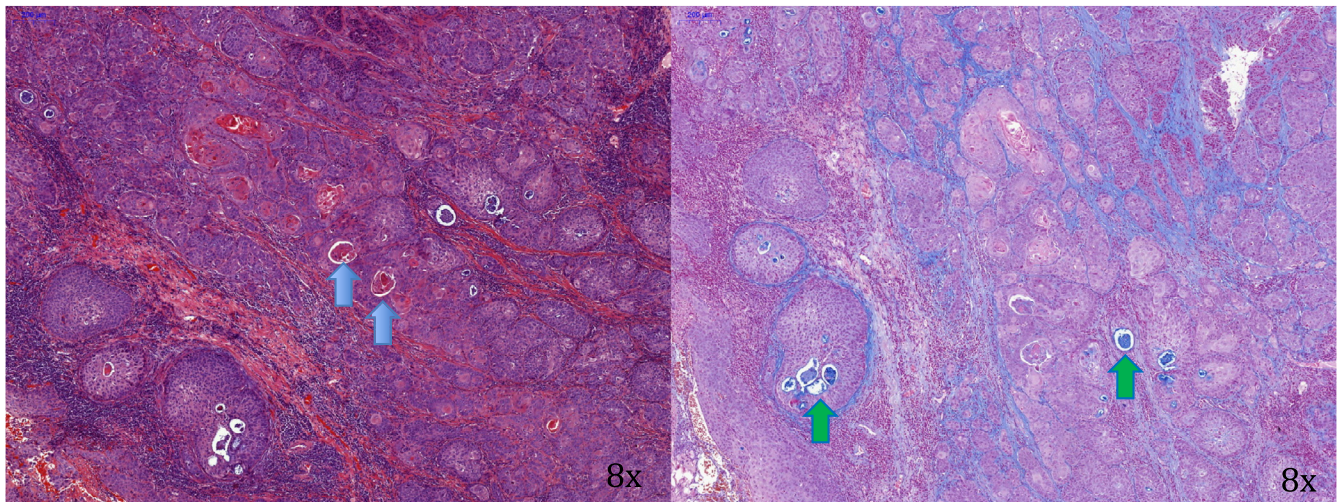


Fig. 2. Biopsy – LEFT (hematoxylin & eosin staining – HE) keratin pearls (blue arrows) and RIGHT (alcian blue – AB) with AB-positive region confirming the secretory nature of the glands (green arrows).

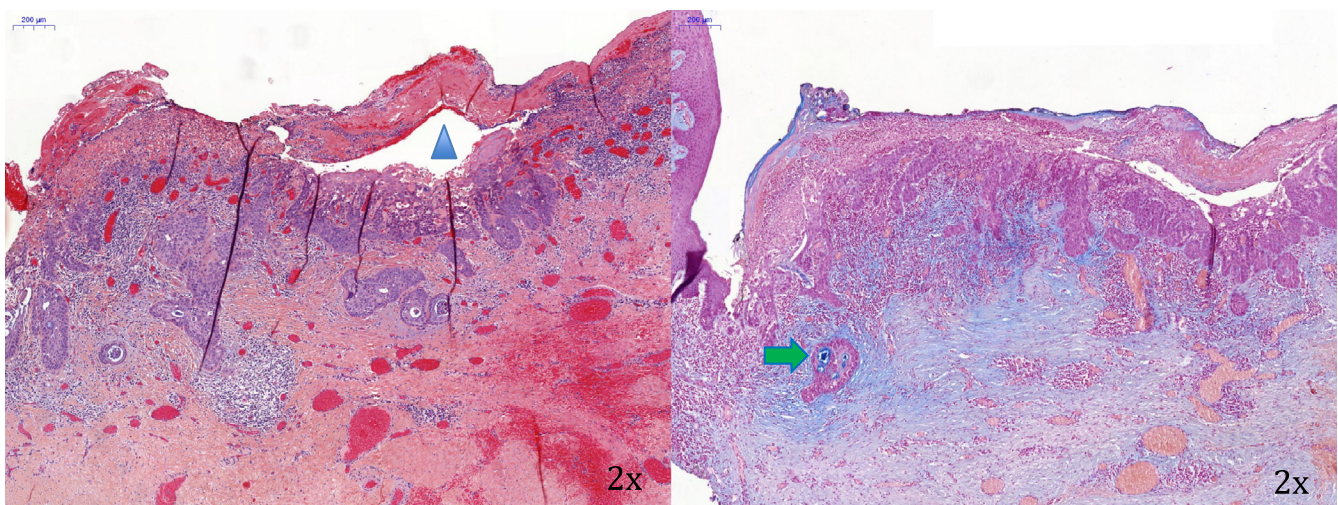


Fig. 3. Surgical piece – LEFT: HE: ulcerated surface (blue triangle) of the tumor and RIGHT: AB: AB-positive in the deeper invasive tumor gland (green arrow).

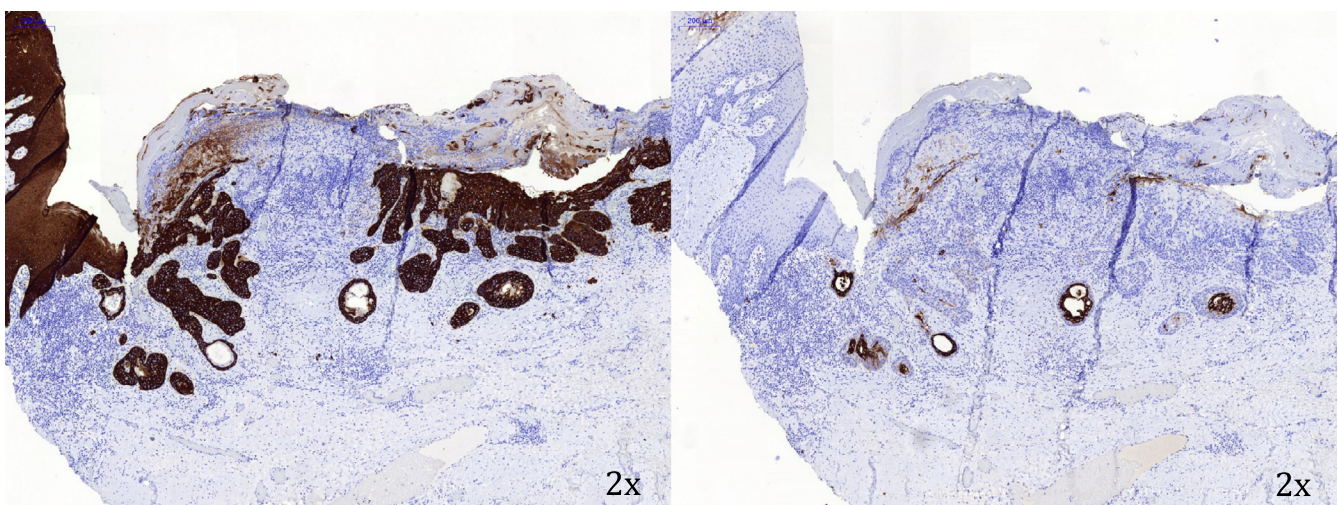


Fig. 4. Surgical specimen – LEFT: CK5/6 positivity for the glandular component and for the epithelial component and RIGHT: CK7 positivity only for the glandular component.

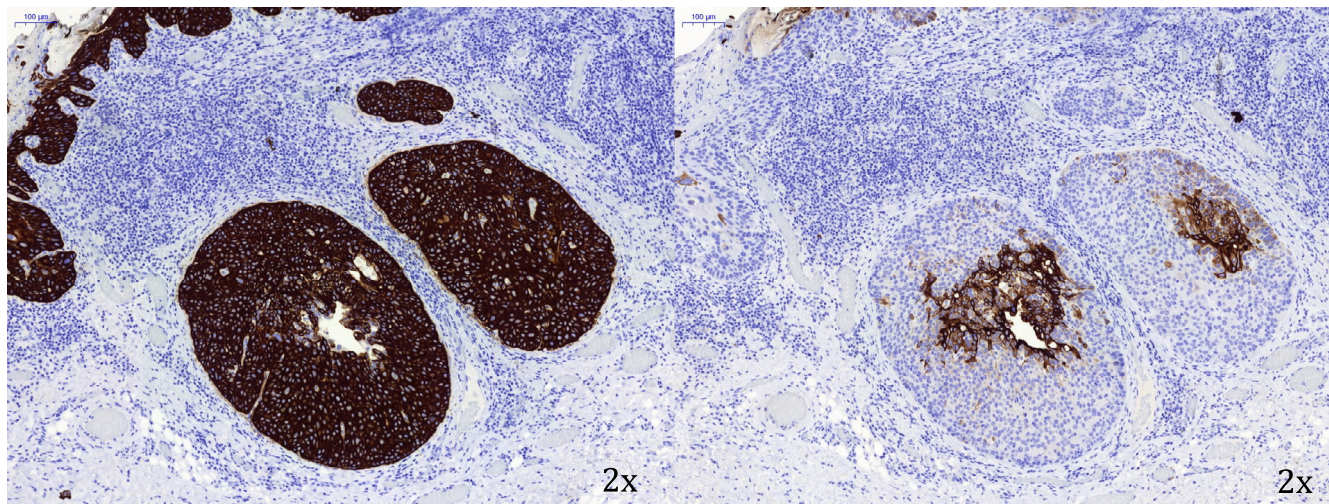


Fig. 5. Surgical specimen – LEFT: CK5/6 and RIGHT CK7 – showing the presence of two distinct cell types.

Table I. Histopathological comparison of ASC and MEC.

Adenosquamous carcinoma	Mucoepidermoid carcinoma
Severe dysplasia or carcinoma <i>in situ</i> of squamous epithelium	No dysplasia of surface epithelium
Squamous origin	Originates in the salivary gland ducts
Keratinization of squamous component	No keratinization
Deep malignant glands	Diffuse glands
Secondary invasion of seromucosal glands	Secondary invasion of squamous epithelium
No lobular arrangement	Lobular arrangement
No intermediate cells	Intermediate cells present

Table II. Histopathological comparison of ASC and ASCC.

	Adenosquamous carcinoma	Adenoid squamous cell carcinoma
Acantholysis	Absent	Present
Glandular structures	Present	Absent
Mucin	Positive (but not a diagnostic criteria)	Negative
Perineural invasion	Frequent	Usually absent
Chronic inflammation	Absent	Usually present

ASCC features both a component-type epidermoid carcinoma and a squamous cell carcinoma on the tumor surface, along with a pseudoglandular part, which corresponds to cells or groups of cells in acantholysis, with cellular debris predominating in these pseudoglandular lumens. The presence of clear cells and inflammatory infiltrate is common (Tab. II). The BSCC is similar in appearance to ASC with PAS or alcian blue-positive squamous or glandular differentiation zones. Nevertheless, BSCC has basaloid solid cell nests with a central comedonecrosis region, which is never present in ASCC.

Conclusion

ASC is a rare tumor of the upper aerodigestive tract that usually progresses rapidly and aggressively. The major differential diagnosis is MEC, ACC, and BSCC.

Conflict of interests: The authors declare that they have no conflicts of interest in relation to this article.

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