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협점막에서 발생한 선편평상피암 증례

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〈Abstract〉

Adenosquamous Carcinoma at the Buccal Mucosa

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Oral adenosquamous carcinoma (ASC) is a rare and aggressive type of squamous cell carcinoma (SCC). It is characterized by a squamous component originating at the mucosa and a deep glandular component. ASC may be misdiagnosed as SCC by superficial incisional biopsy including only the squamous component. ASC has a worse prognosis than general variants of SCC, so accurate diagnosis is essential for patient survival. We present a case report of a large ASC arising in the buccal mucosa, first mistaken as general SCC.

Key words: Adenosquamous carcinoma, Buccal mucosa

Ⅰ. INTRODUCTION

Adenosquamous carcinoma (ASC) is a rare type of squamous cell carcinoma (SCC) composed of two independent histologic components. The squamous component begins at the mucosal epithelium with definite evidence of dysplastic squamous epithelium progressing to SCC. The glandular component locates deep under the squamous component. Glandular component is characterized by a true glandular pattern with mucous, but must not originate from salivary glands or other normal glandular structures[1]. ASC has a predilection for males and elder patients. Most of the case reported of oral ASC occurred in the mouth floor, with a few reports arising in the tongue, palate and mandible[2,3]. In this case report, we described a rare case of ASC arising at the buccal mucosa which was first misdiagnosed as general SCC.

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Ⅱ. CASE REPORT

An 81-year-old male visited the department of Oral and Maxillofacial surgery for oral SCC at the right buccal mucosa. Initial diagnosis was made at another hospital by

incisional biopsy. The patients showed nodular facial swelling at the right upper cheek which had been discovered 20 days before the initial visit at the previous hospital (Fig. 1A). Scar tissue with an irregular ulceration caused by the previous biopsy was observed in the right buccal mucosa (Fig. 1B). Lesional tissue other than the ulcer was not noticed on the buccal mucosa surface. The mass showed tenderness on palpation with pus-like discharge.

Radiologic evaluation by magnetic resonance imaging (MRI) and positron emission tomography-computed tomography (PET-CT) were processed before surgery. MRI images revealed a massive infiltrative tumor at the right buccal mucosa with cystic change in the central (Fig. 1C). Diameter size of the mass exceeded 3.5cm. Tumor infiltration involved right buccinator muscle, distal region of stenson's duct and subcutaneous tissue of the overlying facial skin. A metastasis positive lymph node on right neck at level IB (size 2.2 cm) was seen on the image. PET-CT image showed similar findings to MRI (Fig. 1D).

The incisional biopsy revealed phased dysplasia and carcinoma in situ of the mucosal epithelium leading to invasion (Fig. 1E).

Wide excision of the primary tumor including the skin with right partial maxillectomy, modified radical (right) neck dissection and right total parotidectomy was performed. Reconstruction of the soft tissue defect was done with left radial forearm free flap.

Two distinguishable histological appearances were seen in the tumor by pathological examination (Fig. 2A). A squamous component with moderated differentiation and keratin formation was the main composition. The central area within the squamous component was suspected to be full necrosis and pus on gross examination. Microscopic examination revealed the necrotic area to be neoplastic squamous epithelium and extreme amounts of scattered necrotizing keratin (Fig. 2B). A glandular component was observed at the peripheral of the main squamous component (Fig. 2A). The glandular component had true glandular features composed of columnar and cuboidal neoplastic cells (Fig. 2C). Metastatic cervical lymph nodes with extracapsular extension was noticed in level IB. Extreme extracapsular extension resulted as a mass in the neck masking other possible metastatic lymph nodes. The metastatic foci showed glandular features (Fig. 2D). The pathologic diagnosis was revised to ASC.

After the surgery, the patient was referred to department of Medical Oncology and Radiation Oncology for concurrent chemotherapy and radiation therapy (CCRT).

Ⅲ. DISCUSSION

ASC is aggressive and has a much poorer prognosis than general SCC so early detection and accurate diagnosis is essential for survival[1)]. While oropharyngeal SCC has a 5-year survival rate of about 65%, ASC has a much lower 5-year survival rate of 13% with nearly half of the patient only surviving at a mean duration of 2 years. Cervical lymph node metastasis is more frequent in ASC (about 46%) than SCC (about one-third)[4)]. Although SCC varies in prognosis by location and treatment options, the overall prognosis seems to be worse in ASC than general SCC. The tumor in our case was highly invasive. The tumor portion within the buccal space was larger than 3.5cm while the tumor portion exposed on the mucosal surface was suspected to be about 0.5~1cm estimated by the remaining ulcer and biopsy slides. There were definite evidences of ASC origin at the mucosal surface, so the massive extension to the subcutaneous tissue supported its highly invasive nature. Rapid growth with cystic degeneration and necrotizing keratin was a unique finding in our case. Cystic degeneration is untypical radiologic finding in SCC as well as ASC and may cause confusion during radiologic evaluation.

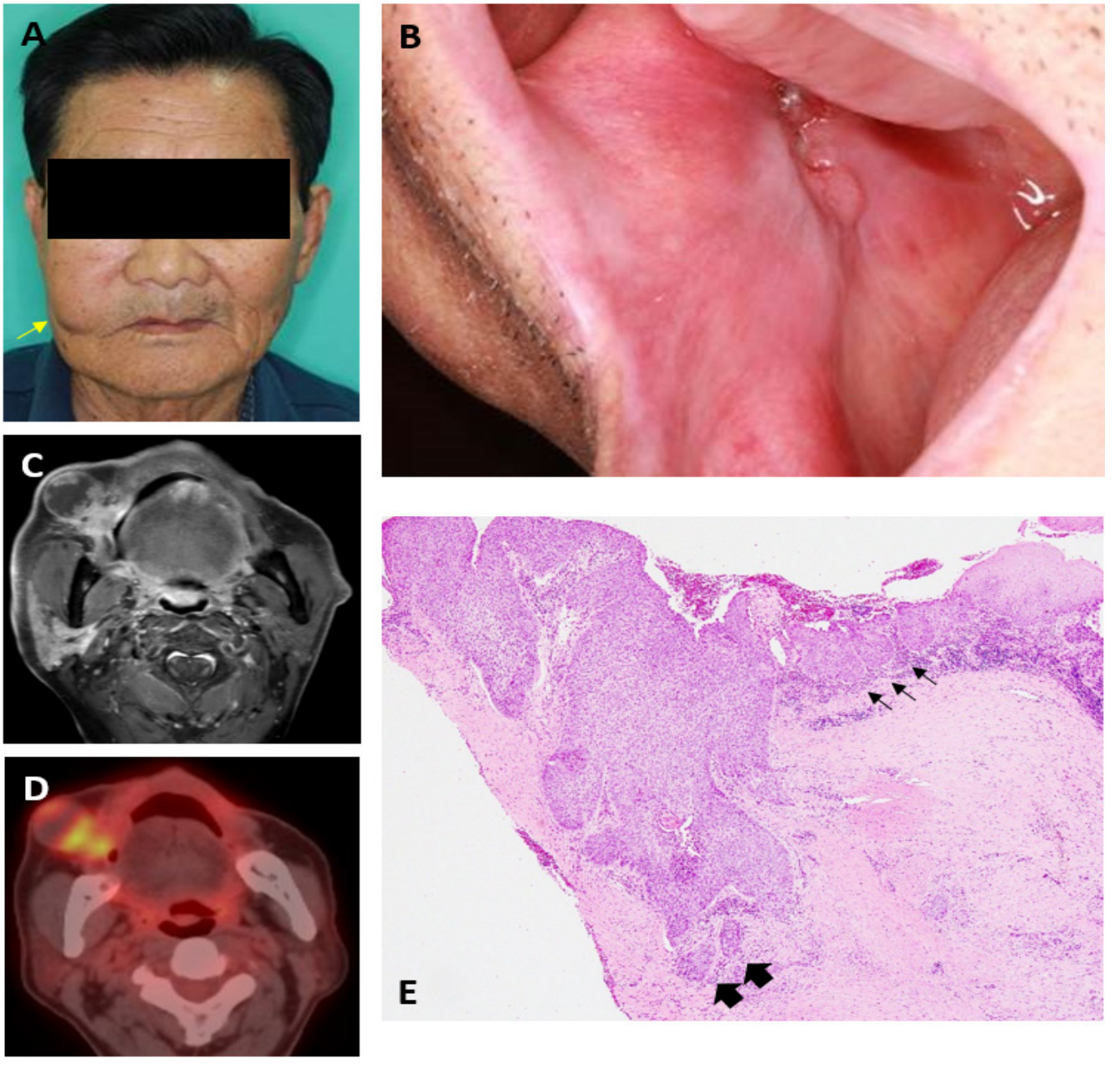

Fig. 1. (A) Extraoral image with facial swelling (yellow arrow) on the right cheek taken on the initial visit at our institute.
(B) An irregular ulceration is seen on oral examination.
(C) Axial magnetic resonance image of the right cheek revealed a large tumor with central cystic degeneration infiltrating from the buccal mucosa to the subcutaneous tissue (T1-weighted image).
(D) Axial positron emission tomography-computed tomography (PET-CT) image of the right cheek with 18F-Fludeoxyglucose (FDG) uptake at the lesion, including the cystic area.
(E) Incisional biopsy of the right buccal mucosa diagnosed as squamous cell carcinoma at the previous hospital. Invasion (thick black arrows) of the mucosal epithelium with adjacent epithelial dysplasia (thin black arrows) confirmed the origin of lesion to be the mucosal surface.

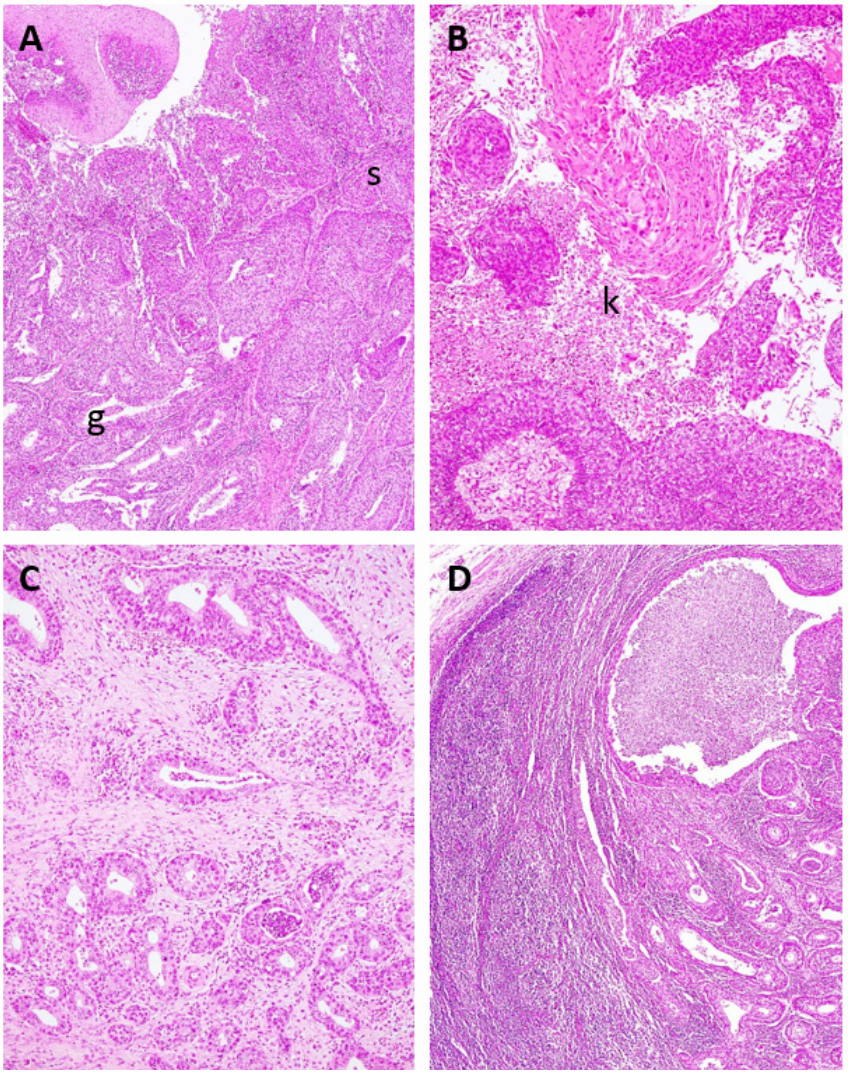

Fig. 2. Histopathologic examination of the surgical specimen.

(A) Two independent components of the tumor. The squamous component [s] was located at the origin and central region of the tumor, while the glandular component [g] was located laterally to the squamous component (H&E staining, ×40).

(B) Squamous component of the cystic region, which showed neoplastic squamous epithelium and necrotizing keratin accumulation [k] (H&E staining, ×100).

(C) Glandular component with true glandular features composed of columnar and cuboidal tumor cells (H&E staining, ×100).

(D) Metastatic foci in the cervical lymph node displayed glandular differentiation (H&E staining, ×40).

ASC may be pathologically mistaken as SCC by superficial biopsy. Even after full excision it needs pathological differentiation with mucoepidermoid carcinoma, adenocarcinoma, and adenoid squamous cell carcinoma[2,5,6]. Mucoepidermoid carcinoma is structured by mucous cell proliferation, intermediate cells and a squamous component that does not originate from the surface epithelium. Adenoid SCC is an entirely true squamous epithelium origin with glandular-like features within the tumor island made by central necrosis due to rapid growth.

ASC arises in mucosal locations that are sufficient of minor salivary glands or openings of major salivary gland[2,3]. The architecture with a superficial squamous component and deeper glandular component resembles the histologic arrangement of a ductal orifice. ASC may have their carcinogenesis origin at salivary gland ductal openings in the oral cavity.

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