

MRI findings of a huge cystadenocarcinoma of the palate

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ABSTRACT

Cystadenocarcinoma of the salivary glands is a very rare, slow growing, and low-grade malignant neoplasm. It is characterized by predominantly cystic growth with or without the intraluminal papillary component. However, it lacks of any additional specific histopathologic features that characterize other types of salivary carcinomas showing cystic growth. Therefore, definite diagnosis of the cystadenocarcinoma is difficult and it is often misdiagnosed. It is conceptually the malignant counterpart of the benign cystadenoma. We present a cystadenocarcinoma on the palate of a 49-year-old man with special emphasis on magnetic resonance imaging. (*Korean J Oral Maxillofac Radiol* 2010; 40 : 191-6)

KEY WORDS : Cystadenocarcinoma; Salivary Glands; Magnetic Resonance Imaging

Cystadenocarcinoma (CAC) is a rare malignant tumor of salivary glands, which has been recently included in the classification schemes of salivary gland tumors.¹ CAC was previously included under other categories, namely “adenocarcinoma, not otherwise specified.” Papillary CAC was first included and named in the 1991 World Health Organization (WHO) classification,¹ and it was reclassified as cystadenocarcinoma in the 2005 WHO classification. However, other synonyms are still being used to describe this entity, such as papillary cystadenocarcinoma,² mucus producing adenopapillary (non-epidermoid) carcinoma,³ malignant papillary cystadenoma,⁴ and low-grade papillary adenocarcinoma of the palate.⁵ About 65% of CACs occur at the major salivary glands, and mostly at the parotid gland.⁶ The buccal mucosa, lips, and palate are the most frequently involved area relating to the minor salivary gland sites.^{6,7} Magnetic resonance imaging (MRI) findings of the palatal CAC have not been reported in previous literature. We report a case of huge CAC in the palate with maxillary bone invasion using by MRI.

Case Report

A 49-year-old man first noted a mass at his palate in 2002, but did not have it treated. He visited Seoul National University Dental Hospital for evaluation and treatment of mobile and

painful right upper and lower molar teeth and the mass of the palate in 2009. Intraoral examination revealed a huge mass with ulceration (Fig. 1). MR imaging revealed enlargement of cervical lymph nodes and metastasis was suggested. Incisional biopsy was performed, with the diagnosis of cystadenocarcinoma.

Panoramic radiograph showed that the posterior wall of the right maxillary sinus, maxillary sinus floor, maxillary tuberosity, and right side of the hard palate were destroyed, therefore suggesting malignant tumor (Fig. 2).

T1 weighted MRI showed a non-homogeneous, low signal intensity mass on the soft palate, obstructing the nasopharynx and lower part of the oropharynx (Figs. 3A and B). T2 weighted MRI demonstrated heterogeneous, low signal intensity with some high signal areas (Fig. 3C). Post-contrast images showed



Fig. 1. Huge mass with ulceration occupies the midpalatal area.

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Fig. 2. Panoramic radiograph shows destruction of the maxillary sinus floor, posterior wall, and hard palate of the right side.

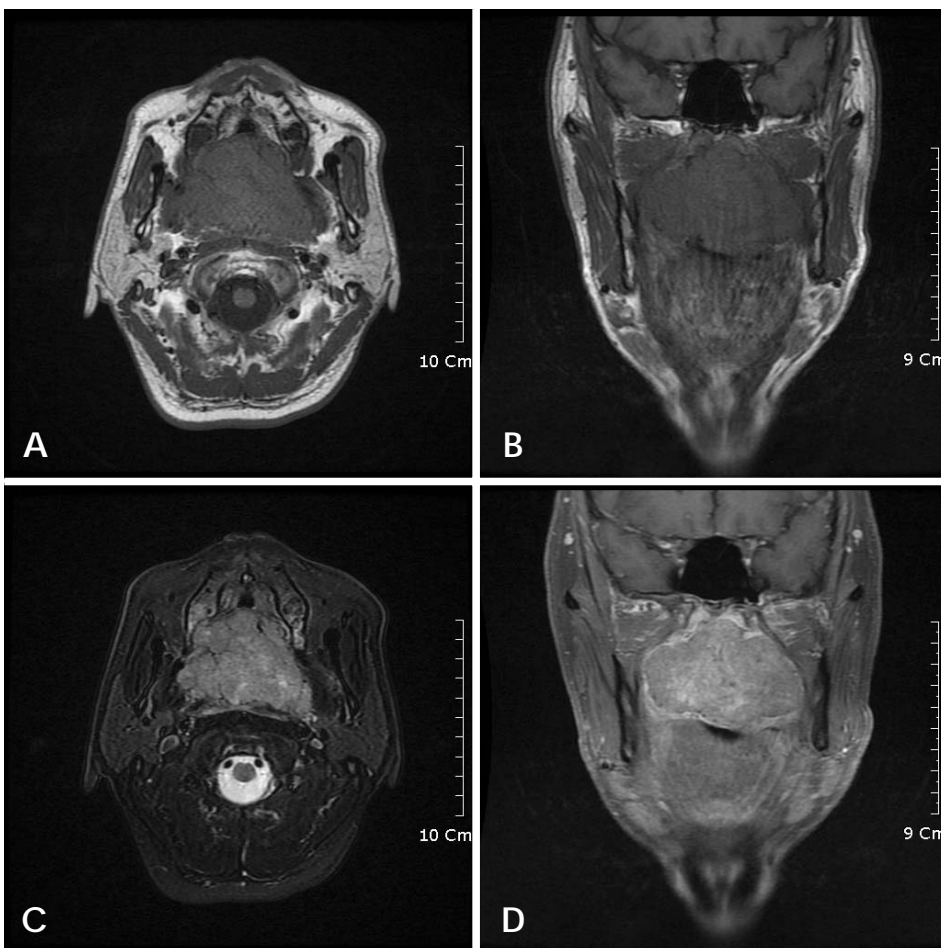


Fig. 3. On T1 weighted images, a heterogeneous, low signal mass obstructs nasopharynx and compresses medial pterygoid muscle on both sides. A. T1 weighted axial image. B. T1 weighted coronal image. C. T2 weighted images showed low signal intensity with some high signal areas. Maxillary invasion of right side shows well-defined margin. D. Post-contrast images show slight enhancement.

slight enhancement (Fig. 3D). Low signal intensity on T2 and slight enhancement suggested high cellularity of the tumor. High signal areas on T2 weighted images which showed no enhancement (Figs. 4A and B), were considered to be necrotic areas. On the left level II area, an enlarged lymph node was

found, with the size reaching 15 mm. Enhancement was shown on the post-contrast images and high uptake on positron emission tomography scan (Figs. 5A-C).

The mass was enormous, invading into the right maxilla and nasal cavity anterolaterally, and extending to the middle nasal

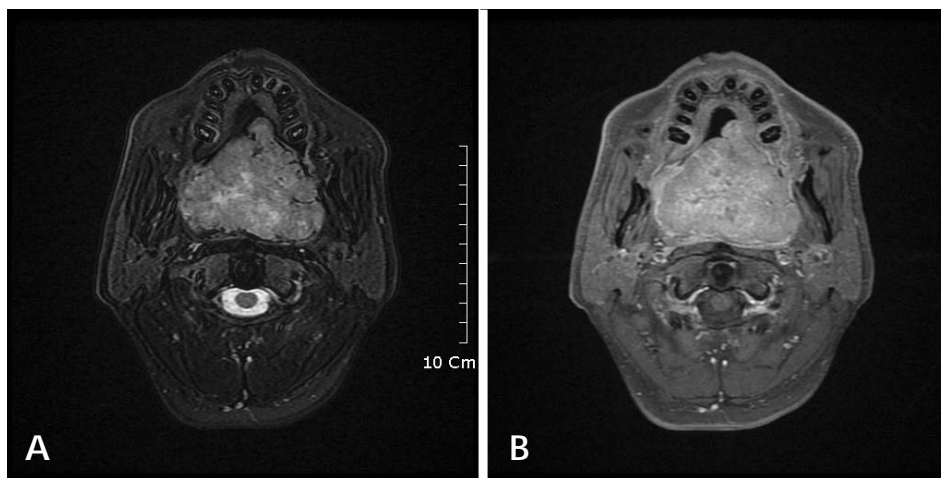


Fig. 4. High signal areas on T2 weighted images show no enhancement. A. T2 weighted image, B. Post-contrast image.

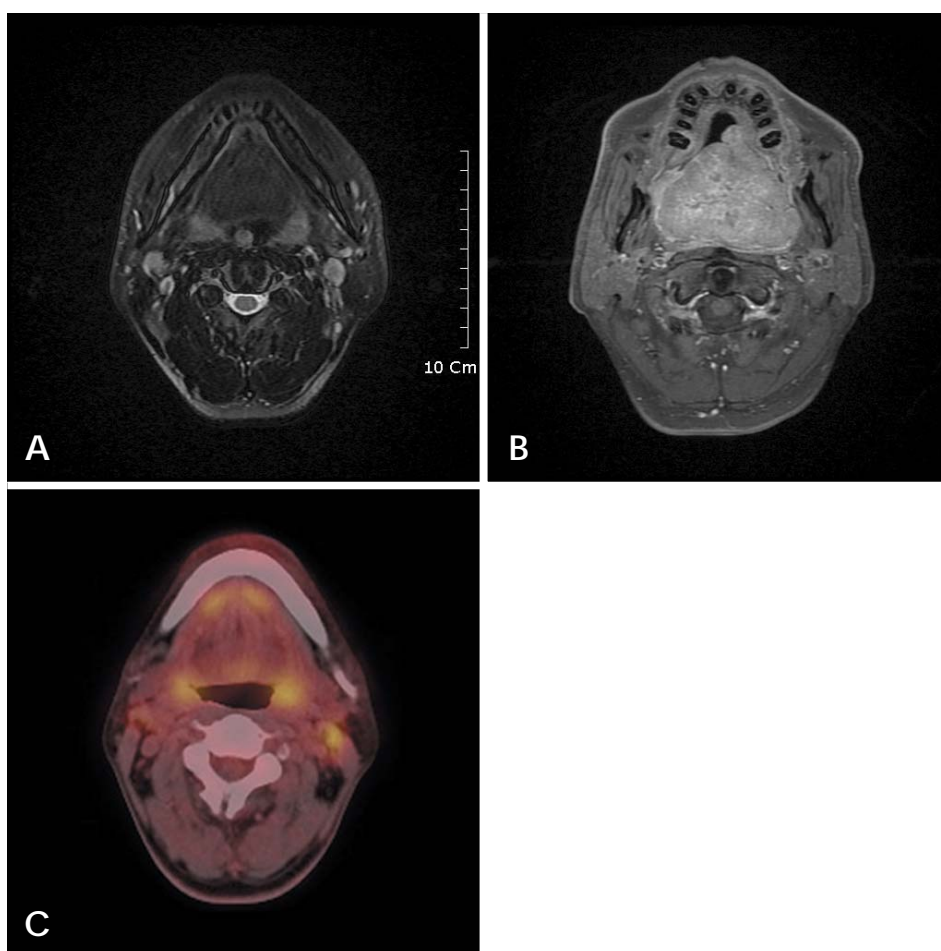


Fig. 5. An enlarged lymph node of left submandibular area shows enhancement on post-contrast images and high uptake on positron emission tomography. A. T2 weighted image. B. Post-contrast image. C. Positron emission tomography.

concha superiorly. It occupied the midpalatal area and medial margins of the medial pterygoid muscle, compressing the muscle on both sides. Also, both pharyngeal walls were displaced.

Complete mass resection with partial maxillectomy and supraomohyoid neck dissection on both sides were performed. Histopathologic examination showed encapsulated mass with

intraluminal papillary growth, low mitotic activity, and low degree of nuclear pleomorphism (Figs. 6A and B). Necrotic areas were also found (Fig. 6C). Metastasis was confirmed for 6 out of 54 regional lymph nodes. Three lymph nodes were on the right level II, one on left level I, and two on left level II (Figs. 7A and B). Therefore it was diagnosed as a low-grade

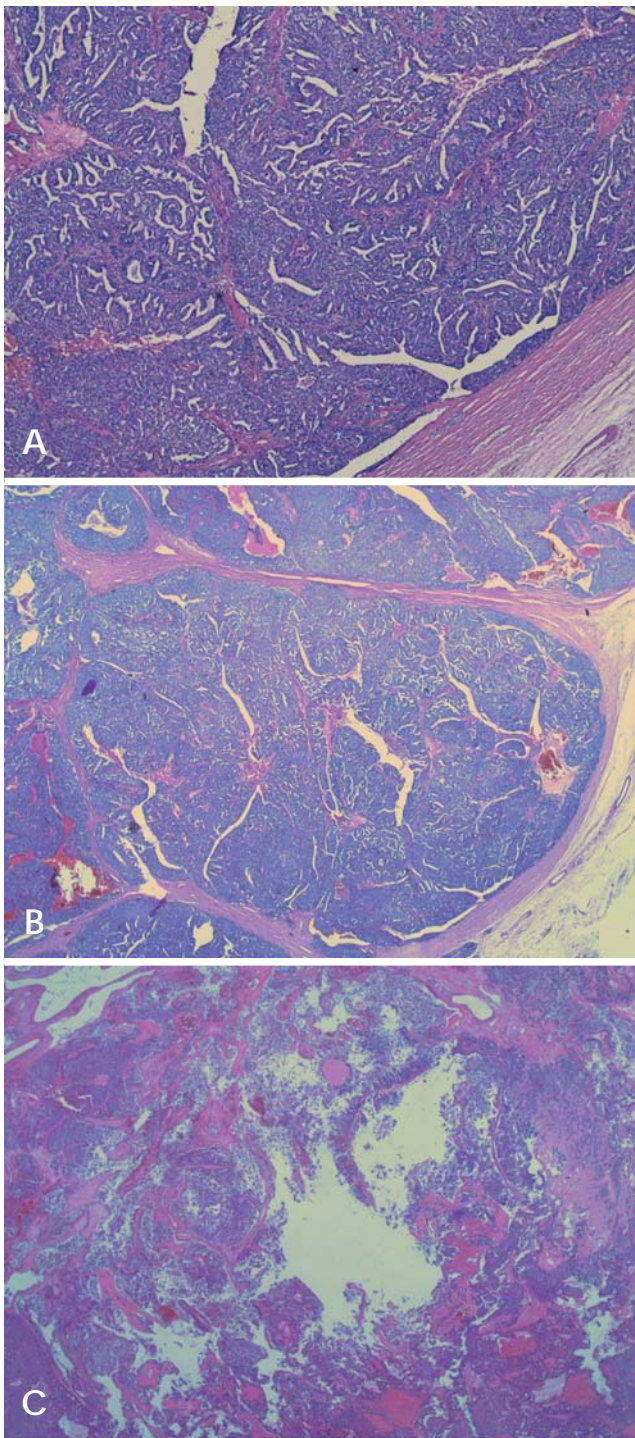


Fig. 6. A. Histologically, the lesion shows fibrous capsule, intraluminal papillary growth, and high cellularity ($\times 40$). B. Mitotic figures are rare and degree of nuclear pleomorphism is low ($\times 400$). C. In the center of slide, necrotic areas are shown ($\times 12.5$).

CAC with papillary formation and metastatic lymph nodes. No recurrence or additional metastatic lymph node was found within a year follow up.

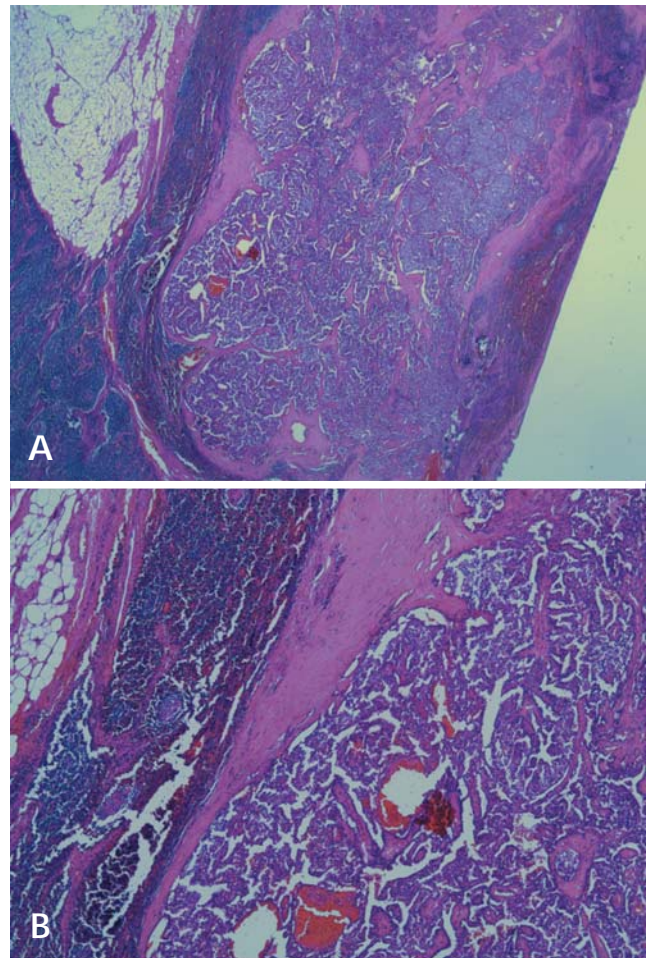


Fig. 7. Tumor cells are invaded into left level II lymph node. A. $\times 12.5$. B. $\times 40$

Discussion

In the 2005 WHO classification, papillary CAC was reclassified as CAC. The word ‘papillary’ was dropped and CAC was defined as “a rare malignant tumor characterized by predominantly cystic growth which lacked any additional specific histopathologic findings that characterized other types of salivary carcinomas showing cystic growth”.

Foss et al.⁶ also mentioned that “papillary formations may or may not be prominent and do not influence their behavior”. In their study on a total of 57 cases of CACs in 1996, no sex predilection could be seen. The average age of patients was 59 years; more than 70% were over 50 years of age. The mass size ranged from 0.4 to 6.0 cm in maximum dimension. The average size of the tumors were 2.2 cm in the minor glands and 2.4 cm in the major glands.⁶ In our case, since the patient had ignored the lesion for 7 years, the resected tumor had grown to a size of 7.5 \times 7.0 \times 5.0 cm (Figs. 8A and B).

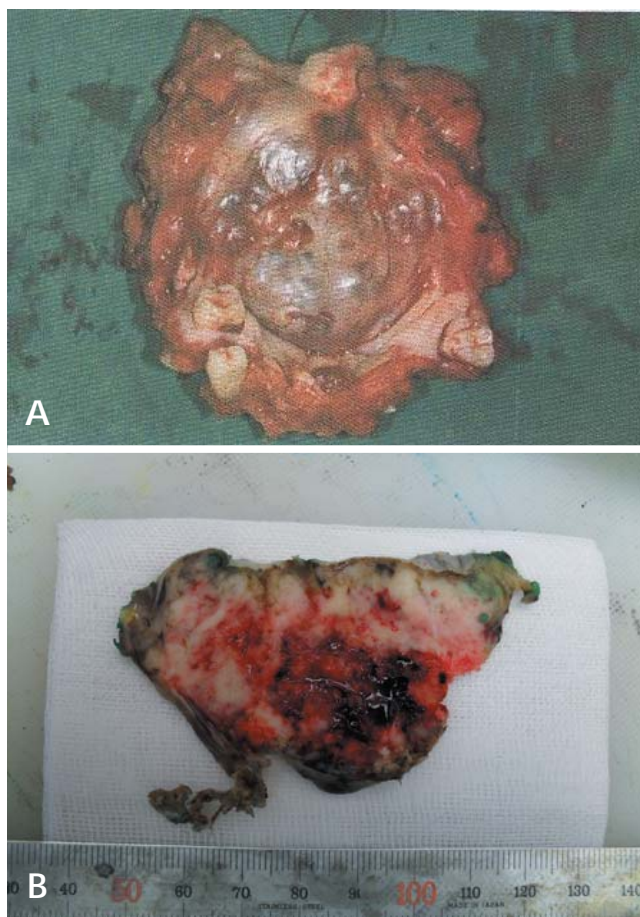


Fig. 8. A. The complete mass with partial maxillectomy. B. Resected tumor is so huge, reaching the size of $7.5 \times 7.0 \times 5.0$ cm.

Only a few MRI findings of CACs in the minor salivary gland have been reported in the literatures, with insufficient information and no specific findings.⁸⁻¹¹ Harimaya et al.⁸ reported a CAC on the submandibular gland and it showed low signal intensity on T1 and intermediate to high signal intensity on T2 weighted images. There was no information of enhancement. Tomioka et al.⁹ reported a CAC at the mouth floor and it showed slightly elevated signal intensity on T1 and high signal intensity on T2 weighted images. There was no information about enhancement also either. Nakagawa et al.¹⁰ reported a CAC of tongue, which showed enhancement on the outer surfaces of the mass. However, no additional description was given. Koç et al.¹¹ reported a CAC on the submandibular gland and it showed non-homogenous hypointensity on T1 and heterogeneous hyperintensity on T2 weighted images. Also enhancement of the solid part was reported.

In this case, non-enhanced but high signal areas on T2 weighted images turned out to be necrotic areas by histopathologic examination (Fig. 6C). Tumors with low signal intensity

on T2 weighted images and slight enhancement usually meant having high cellularity.¹²⁻¹⁴ As expected, the tumor showed high cellularity (Figs. 6A and B)

The tumor had destroyed the right maxilla, however the bone-tumor interface was smooth and well-defined (Fig. 3C). It might be because the tumor has low-grade malignancy and was encapsulated by fibrous capsule (Figs. 6A and B). Interestingly, although the tumor had low-grade malignancy and well defined margin with fibrous capsule, metastatic lymph nodes were found. In a study of 40 patients with follow-up data, CAC behaved as a low-grade manner.⁶ All patients were free of tumor within a mean interval of 59 months after their initial surgery, and either alive (36 patients) or dead from other causes (four patients). Because CACs showed a low-grade malignancy, CT scan or MRI might not always reveal the typical characteristics of malignant tumor.^{11,15} However, there were some reported cases that had an unexpected aggressive behavior and lymph nodes metastasis.^{6,15-17}

Even with the evaluation of preoperative imaging, it may be difficult to make a definite diagnosis of CAC.^{2,15} Definite diagnosis of CAC is essential in making a treatment plan. Therefore comprehensive histopathological assessment by incisional or fine needle aspiration biopsy is required for diagnosis. MR imaging is also important for confirming the presence of a mass, allowing evaluation of the extent and outline of the tumor, along with the relationship to the adjacent structure and lymph nodes metastasis.

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