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Basal cell adenocarcinoma in the retromolar trigone: A case report

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ABSTRACT

Basal cell adenocarcinoma, considered to be the malignant counterpart of basal cell adenoma, is a rare, low-grade malignant tumor of the salivary glands, accounting for 1-2% of salivary gland malignancies. It predominantly affects the parotid gland, while involvement of the minor salivary glands is exceptionally rare. This report presented a case of basal cell adenocarcinoma involving the left retromolar trigone in a 54-year-old woman. The initial provisional diagnosis suggested a benign or low-grade malignant salivary tumor. Advanced magnetic resonance imaging techniques, including diffusion-weighted imaging and apparent diffusion coefficient analysis, aided in the preoperative prediction of malignancy, and an incisional biopsy confirmed the diagnosis of basal cell adenocarcinoma. This case underscored the challenge of distinguishing basal cell adenocarcinoma from benign salivary tumors, as clinical and imaging features often overlap. Surgical excision remains the primary treatment, yielding favorable outcomes; however, long-term follow-up is crucial due to the risk of recurrence. ***(Imaging Sci Dent 2025; 55: 96-101)***

KEY WORDS: Adenocarcinoma, Basal Cell; Salivary Gland, Minor; Magnetic Resonance Imaging

Basal cell adenocarcinoma is a rare malignant tumor of the salivary glands, accounting for 1-2% of salivary gland malignancies, and most frequently occurs in adults in their seventh and eighth decades of life.[1] It most commonly arises in the parotid gland (71.7%), followed by the submandibular gland (7.1%), and is rarely found in the minor salivary glands.[2] While most cases arise *de novo*, some develop from preexisting basal cell adenomas.[3]

The clinical presentation of basal cell adenocarcinoma often includes a slow-growing, painless mass in the affected gland. Because of its indolent growth, it may initially be mistaken for a benign lesion, thereby delaying diagnosis and treatment.[1,4] Computed tomography (CT) and magnetic resonance imaging (MRI) are useful for assessing the tumor's extent and its relationship with surrounding structures; however, definitive diagnosis relies on histopathological and immunohistochemical analysis.[3,5] Histologically, the tumor is characterized by a proliferation of basaloid cells arranged in trabecular, tubular, or solid patterns, often with peripheral palisading. In contrast to basal cell adenomas, basal cell adenocarcinoma exhibits infiltrative growth, perineural invasion, and, in rare cases, distant metastasis.[4] Nevertheless, its biological behavior is generally less aggressive than that of other high-grade salivary malignancies, leading to relatively favorable outcomes when adequately treated.[2,5]

The management of basal cell adenocarcinoma typically involves surgical excision with clear margins, which remains the cornerstone of treatment. The use of adjuvant therapies, such as radiotherapy, is debated and is typically reserved for cases with high-risk features, including positive margins, perineural invasion, or recurrent disease.[3-6] Long-term follow-up is essential due to the potential for late recurrence or metastasis, even when initial treatment appears adequate.[4]

Basal cell adenocarcinoma involving the minor salivary glands is exceedingly rare, and no cases originating in the retromolar trigone have been reported. The purpose of this report is to present a rare case of basal cell adenocarcinoma in the retromolar trigone and to identify clinical

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or imaging features that might distinguish it from benign salivary gland tumors.

Case Report

This study received approval from the Institutional Review Board of Pusan National University Dental Hospital in Yangsan, Korea.

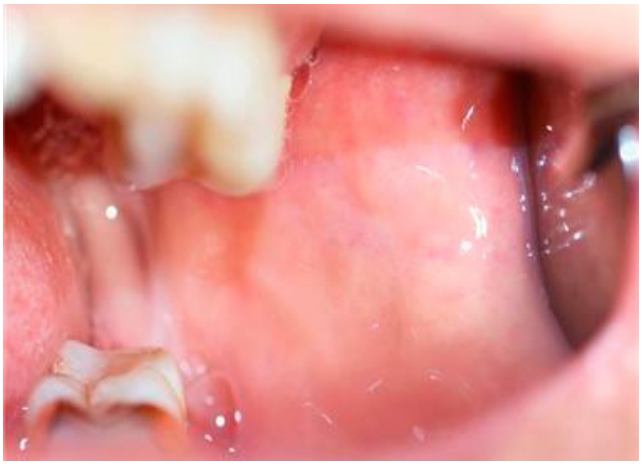

Fig. 1. An intraoral examination reveals slightly swollen mucosa, showing a normal-colored, smooth surface in the left retromolar trigone.

A 54-year-old female patient presented with swelling in the left retromolar trigone. She first experienced discomfort at the site three months prior and reported slight discomfort when opening her mouth. The patient had no significant personal or family history. Intraoral examination revealed mildly swollen mucosa with normal color and texture (Fig. 1). Palpation indicated that the mass was generally soft, with areas of firmer consistency. Panoramic radiography and CT imaging obtained at the initial visit revealed pressure-induced bone resorption of the maxillary tuberosity and anteromedial ramus due to compression by a soft tissue tumor measuring $21 \times 14 \times 16$ mm. The cortical bone remained intact, and no enlarged cervical lymph nodes were observed (Fig. 2). The provisional diagnosis favored either a benign or low-grade malignant salivary gland tumor. Although benign tumors such as pleomorphic adenoma could not be entirely ruled out, the tumor was considered malignant given its origin in the minor salivary gland. Differential diagnoses included mucoepidermoid carcinoma—previously reported in the retromolar trigone,[7] adenoid cystic carcinoma, which occurs relatively frequently, and adenocarcinoma. However, the tumor's small size and lack of distinctive features compli-

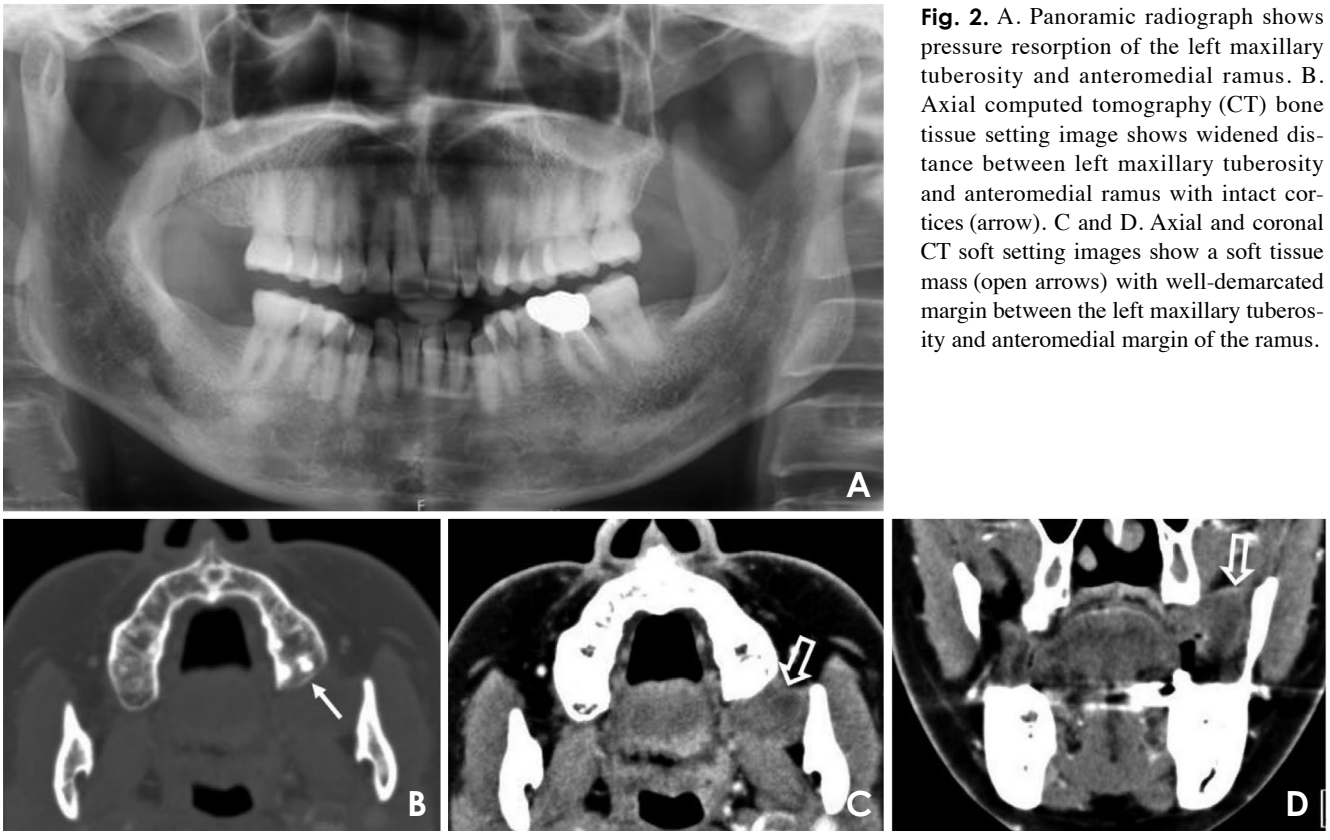

Fig. 2. A. Panoramic radiograph shows pressure resorption of the left maxillary tuberosity and anteromedial ramus. B. Axial computed tomography (CT) bone tissue setting image shows widened distance between left maxillary tuberosity and anteromedial ramus with intact cortices (arrow). C and D. Axial and coronal CT soft setting images show a soft tissue mass (open arrows) with well-demarcated margin between the left maxillary tuberosity and anteromedial margin of the ramus.

cated the differential diagnosis. Therefore, an incisional biopsy was performed approximately 2 months after the initial visit and revealed basal cell adenocarcinoma. Approximately 1 month after the biopsy (three months after the initial visit), additional clinical photography, computed tomography (CT), magnetic resonance imaging (MRI), and positron emission technology-CT scans were obtained. The oral examination now revealed a fixed, ulcero-proliferative mass that had increased in size compared to the initial assessment (Fig. 3). CT imaging showed that the tumor now measured $28 \times 22 \times 18$ mm, with cortical resorption of the maxillary tuberosity, and displacement of the medial pterygoid muscle (Fig. 4). MRI revealed a well-marginated mass lesion that exhibited low signal intensity on T1-weighted images, hypo- to intermediate signal intensity on T2-weighted images, and heterogeneous enhancement on gadolinium-enhanced T1-weighted images. Diffusion restriction on the apparent diffusion coefficient (ADC) map indicated limited water diffusion—a feature often seen in regions of high cellular density that suggests malignancy. Accordingly, diffusion-weighted imaging (DWI) was performed, and the ADC value was measured. DWI ($b = 1000$ s/mm^2) demonstrated a hyperintense mass, and the ADC map revealed significant diffusion restriction with an ADC value of 0.91×10^{-3} mm^2/second (Fig. 5).

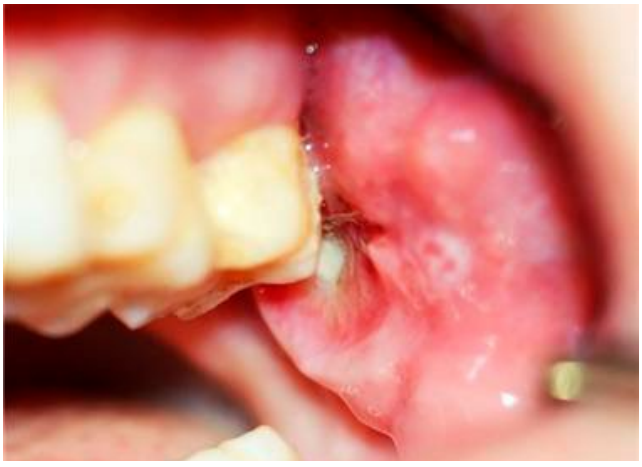

Fig. 3. Intraoral examination reveals a bumpy swollen ulcerated mucosa due to damage from the opposing tooth.

The patient underwent wide excision with selective neck dissection. Histopathological examination using hematoxylin and eosin staining revealed tumor islands and strands infiltrating the surrounding tissues. The tumor was poorly demarcated, lacked encapsulation, and exhibited perineural invasion. It was composed of both basaloid and ductal cells, with a thin, eosinophilic, hyalinized stroma surrounding the tumor nests. The neoplastic clusters contained two distinct cell types: central polygonal pale-staining cells and peripherally arranged hyperchromatic cells. The peripheral cells, which had scant cytoplasm, displayed a palisading arrangement (Fig. 6). The cervical and submandibular lymph nodes were free of tumor involvement, confirming the diagnosis of basal cell adenocarcinoma. The patient was followed for 2 years without evidence of recurrence or metastasis.

Discussion

The term "basal cell adenocarcinoma" was first introduced in 1990 by Ellis and Wiscovitch,[8] who described a major salivary gland epithelial tumor that exhibited the clinicopathologic characteristics of basal cell adenoma but also demonstrated malignant potential through infiltrative, perineural, and intravascular growth patterns, as well as occasionally increased mitotic activity. In 1991, the WHO classified this tumor as basal cell adenocarcinoma, and by

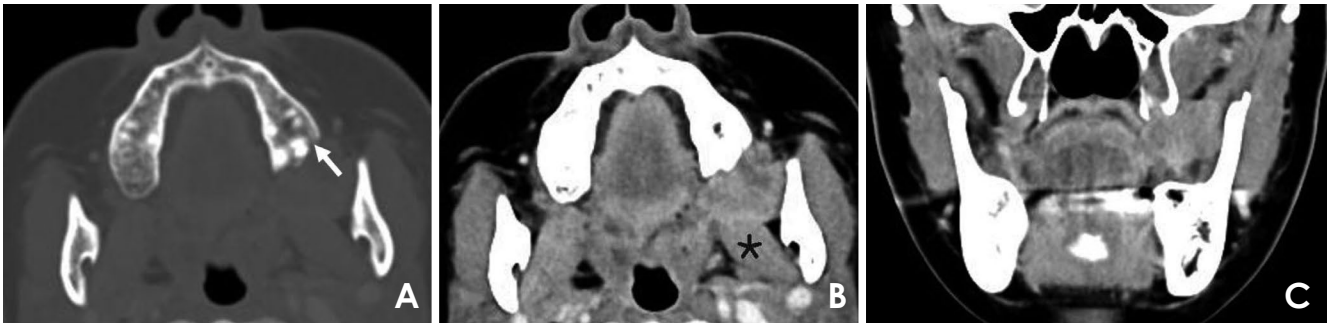

Fig. 4. A. Axial computed tomography (CT) bone tissue setting image shows cortical bone erosion (arrow) of the left maxillary tuberosity. B and C. Axial and coronal CT soft setting images show a soft tissue mass that has grown larger compared to 3 months ago and is pressing on the mesial pterygoid muscle (asterisk).

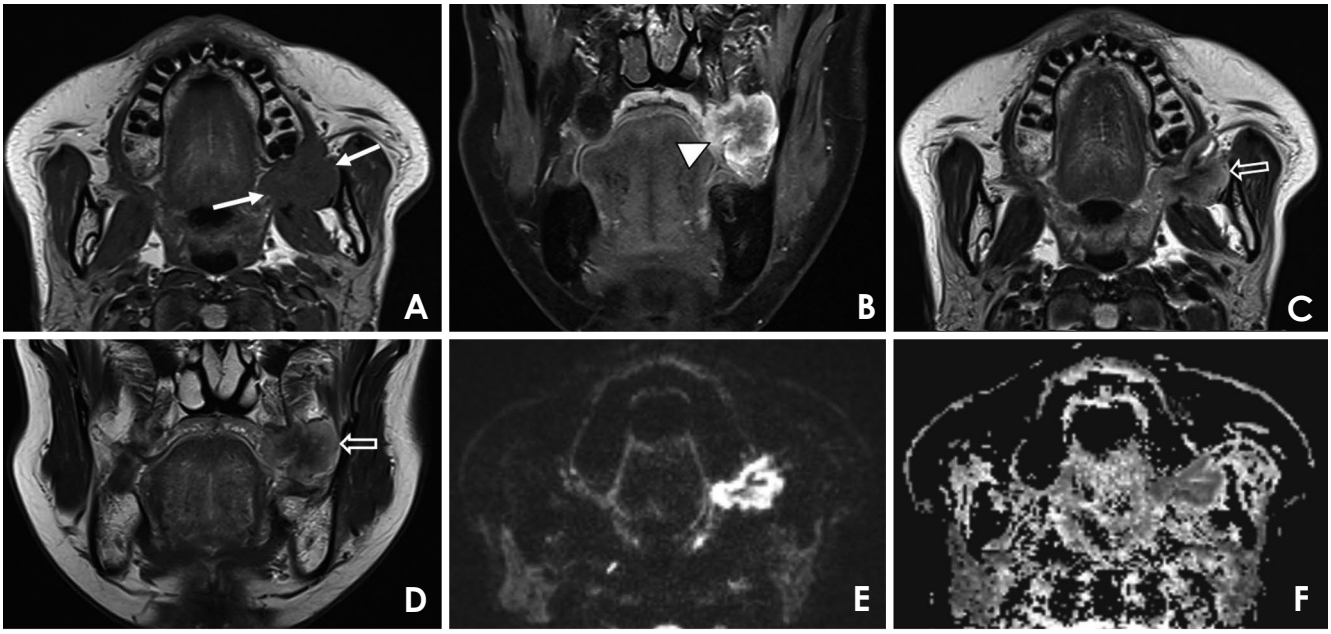

Fig. 5. A. Axial T1-weighted magnetic resonance image shows a tumor (arrows) with low signal intensity. B. Enhanced coronal T1-weighted image shows a tumor with marginal enhancement (arrowhead). C and D. Axial and coronal T2-weighted images show a heterogeneously enhancing mass (open arrows). E. Diffusion-weighted image ($b = 1000$ s/mm^2) shows the tumor to be hyperintense, indicating retracted diffusion. F. Corresponding apparent diffusion coefficient map reveals hypoinsense mass meaning significant restriction.

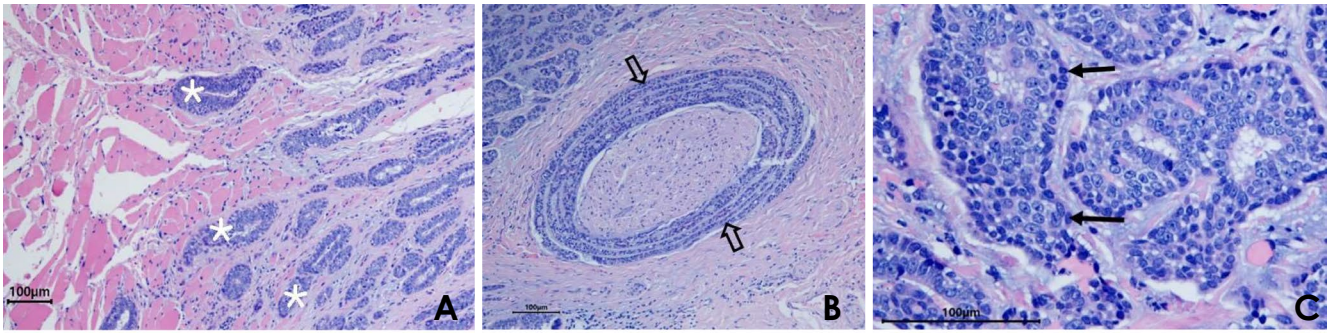

Fig. 6. A. Unencapsulated tumor nests (asterisks) invade adjacent tissue (×100, H&E statin). B. Perineural invasion by tumor cells (open arrows) is shown (×100, H&E statin). C. Biphasic tumor of tubular pattern with central ductal cells and peripheral basaloid cells is seen. Basaloid cells (arrows) with scant cytoplasm and hyperchromatic ovoid nuclei show a palisading arrangement. The tumor shows a few mitotic figures (×400, H&E statin).

2005, the WHO definition emphasized its nature as an infiltrative epithelial neoplasm.

Basal cell adenocarcinoma occurs across a broad age range, most commonly affecting individuals in their sixth to seventh decades, with no clear sex predilection.[1,2,3,9,10] It primarily occurs in the major salivary glands, particularly the parotid gland, and is very rare in the minor salivary glands.[1,3] Cuthbertson et al.[6] reported that in the minor salivary glands, basal cell adenocarcinoma most frequently arises in the palate (32%), followed by the buccal mucosa (18%), nasal cavity and/or sinuses (10%), floor of the mouth (7%), and upper lip (7%). Distinguishing between benign and low-grade malignant salivary gland tumors is challenging, as no significant differences in sex, age, tumor diameter, or location have been observed between basal cell adenocarcinoma and basal cell adenoma cases.[11] Basal cell adenocarcinoma usually presents as an asymptomatic mass with an average size ranging from 2 to 4 cm.[4,6,10] There are no specific clinical or imaging features that clearly differentiate it from basal cell adenoma. In our case, only slight swelling and mild tenderness were observed clinically, and CT imaging revealed a well-defined, low-density

tumor, making it difficult to distinguish between malignant and benign lesions.

Contrast-enhanced MRI is considered the optimal modality for evaluating salivary gland tumors. Imaging features such as low T2 signal, irregular margins, extraglandular spread, cervical lymphadenopathy, and perineural invasion suggest malignancy, although overlap exists between benign and malignant appearances.[11] Recently, advanced MRI techniques that combine diffusion-weighted imaging and perfusion MRI have aided in distinguishing benign from malignant salivary gland tumors.[11,12] Moreover, MR diffusion imaging with voxel-based quantitative analysis using ADC measurements has enhanced our understanding of the various histopathological grades of salivary tumors.[13] In general, malignant tumors lacking cystic or necrotic components tend to exhibit lower average ADC values than benign tumors, likely due to increased cellular density and a higher nucleus-to-cytoplasm ratio.[14] Eida et al.[15] reported that areas with extremely low ADC values were more common in malignant tumors. Although Habermann et al.[16] cautioned that there is considerable overlap in ADC values, studies have shown that benign tumors typically have significantly higher ADC values than malignant tumors, enabling ADC maps to help predict tumor nature preoperatively.[14] Habermann et al.[16] further noted that pleomorphic adenomas could be differentiated from malignant tumors—including mucoepidermoid carcinomas, acinic cell carcinomas, and basal cell adenocarcinomas—based on their higher average ADC values. However, Warthin's tumors often exhibit ADC values similar to malignant tumors and cannot be distinguished on this parameter alone. In one study, the mean ADC values for pleomorphic adenoma, Warthin's tumor, and basal cell adenocarcinoma were 2.09, 0.89, and 0.96 ($\times 10^{-3}$ mm^2/s), respectively. In our case, low signal intensity on T2-weighted images, diffusion restriction on DWI, and an ADC value of 0.91×10^{-3} mm^2/s were indicative of malignancy.

Histopathologically, basal cell adenocarcinomas share features with basal cell adenomas. The critical distinction is the infiltrative growth into surrounding tissues observed in basal cell adenocarcinoma.[1,4,5] Additionally, perineural or vascular invasion and sparse mitotic activity are features of basal cell adenocarcinoma that are absent in basal cell adenoma.[1,9,17,18] In our case, infiltrative growth, perineural invasion, and a few mitoses were observed. The accepted treatment for basal cell adenocarcinoma is complete surgical excision, often followed by adjuvant radiotherapy.[2,4,19] Many studies indicate that basal cell adenocarcinoma is a low-grade malignancy with a generally favorable prognosis.[1,3,18] Although local recurrence occurs in up to 50% of cases, regional lymph node and distant metastases—and death from the disease—are rare.[3,6,18] However, for basal cell adenocarcinoma of the minor salivary gland, Cuthbertson et al.[6] reported a mortality rate of 11%, while Rito et al.[4] reported that 12 out of 22 patients (54.5%) died. Overall, while basal cell adenocarcinoma generally has a good prognosis, unfavorable outcomes do occur, necessitating long-term follow-up after treatment.

In conclusion, this report presented a rare case of basal cell adenocarcinoma of the minor salivary glands arising from the retromolar trigone. Diagnosing basal cell adenocarcinoma is challenging because clinical and imaging evaluations may not clearly distinguish it from other benign and malignant salivary gland tumors. Preoperative prediction of tumor malignancy is crucial, as it significantly influences the surgical strategy. Although advanced MRI techniques - including DWI and ADC analysis - are not specific to any single diagnosis, they provide valuable information regarding the tumor's benign or malignant nature. Surgical excision remains the primary treatment modality. Despite its classification as a low-grade tumor with a generally favorable prognosis, long-term follow-up is necessary.

Conflicts of Interest: None

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