

Basal Cell Adenoma Presenting as a Parapharyngeal Space Mass: A Case Report¹

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Basal cell adenoma is a rare benign epithelial neoplasm of the salivary gland that occurs most frequently in the parotid glands. However, there have been few reports on basal cell adenoma arising from the minor salivary glands in the parapharyngeal space. Here we report a case of basal cell adenoma presenting as a parapharyngeal mass in a 35-year-old woman.

Index words : Salivary gland, neoplasms

Pharynx

Head and neck

Basal cell adenoma is an uncommon benign neoplasm of the salivary gland, accounting for 2% of all primary salivary gland tumors. Histologically, it consists of monomorphic basaloid epithelial cells without a myxochondroid stromal component, which distinguishes it from the pleomorphic adenoma (1, 2). It occurs most frequently in the parotid gland, uncommonly in the oral cavity, upper and lower lip, hard palate, and submandibular gland (3).

There have been few reports of basal cell adenoma occurring in the parapharyngeal space (3 - 5). Here we report a case of basal cell adenoma presenting as a parapharyngeal mass that was incidentally detected.

Case Report

A 35-year-old woman visited our institute with a complaint of headache and dizziness and a brain MRI was

performed. On MR imaging, a portion of a well-circumscribed solid mass was incidentally found in the left parapharyngeal space (Fig. 1A - C). The patient was transferred to an otorhinolaryngologic specialist for evaluation and treatment of the parapharyngeal mass. The patient had no subjective symptoms for the parapharyngeal mass. Furthermore, the mass could not be palpated on physical examination.

For further evaluation of the mass, a neck CT scan was performed and showed a 3×2×3 cm sized, well-defined, inhomogeneously enhanced solid mass in the left parapharyngeal space at the level of the naso-oropharynx. The mass was confined to the parapharyngeal space, obliterating the parapharyngeal fat, and was separated from the deep lobe of the parotid gland by a fat plane and displaced the internal carotid artery posterolaterally (Fig. 2A, B). There were no abnormally enlarged lymph nodes in both sides of the neck. We suggested the possibility of a pleomorphic adenoma arising from minor salivary gland or a schwannoma to a clinician.

The mass was extirpated by the transcervical approach below the mandibular angle and was easily separated from the deep lobe of the parotid gland. On gross pathological examination, the mass was yellowish white and palpated hard. Hemorrhage or necrosis in the mass

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Received December 27, 2006 ; Accepted March 19, 2007

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was not revealed. On histological examination, the tumor cells revealed eosinophilic cytoplasm, indistinct cell borders and round to oval nuclei, distributed in tubular and trabecular patterns with prominent peripheral palisading (Fig. 3A). Immunohistochemical staining for cytokeratin was positive in the central portion of the tumor cell nests, which is indicative of ductal epithelial differentiation (Fig. 3B) and staining for smooth muscle actin was positive in the palisading cells at the periphery of the tumor cell nests, which is indicative of myoepithelial differentiation (Fig. 3C). Staining for GFAP (glial fibrillary acidic protein) was negative in the tumor cells. The final diagnosis was a basal cell adenoma, and the tumor was considered to have originated in the minor salivary gland in the parapharyngeal space.

Discussion

Salivary gland tumors represent less than 3% of all neoplasms of the head and neck. Basal cell adenoma is one of the rare, benign salivary gland tumors, accounting for only 2% of all tumors arising from the salivary glands and is mainly located in the parotid gland (3).

Basal cell adenoma originally was classified as part of the category of monomorphic adenomas, which is an obsolete term. However, basal cell adenoma was recognized as a histological distinct entity in 1991 by the World Health Organization (WHO) (1 - 3). Histologically, it is a benign tumor composed of basaloid cells organized with a prominent basal cell layer, and is sharply delineat-

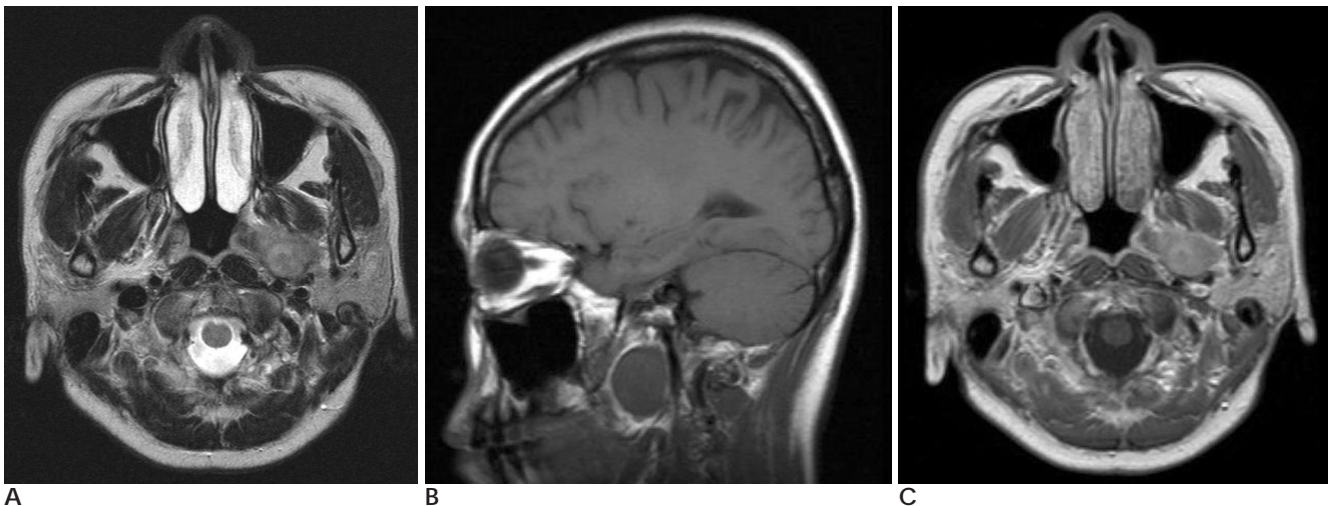


Fig. 1. A 35-year-old woman presented with headache and dizziness; routine brain MR imaging was performed.

A - C. Axial T2-weighted (A) and sagittal T1-weighted (B) images show a well circumscribed, round mass in the left parapharyngeal space, displacing the internal carotid artery posteriorly. On a contrast-enhanced T1-weighted image (C), the mass is inhomogeneously enhanced.



Fig. 2. A, B. A contrast-enhanced axial (A) and coronal (B) CT scan show 3 × 2 × 3 cm sized, inhomogeneously enhanced solid mass with a central low density area. The mass obliterates the parapharyngeal fat and is separated from the deep lobe of the parotid gland by a fat plane.

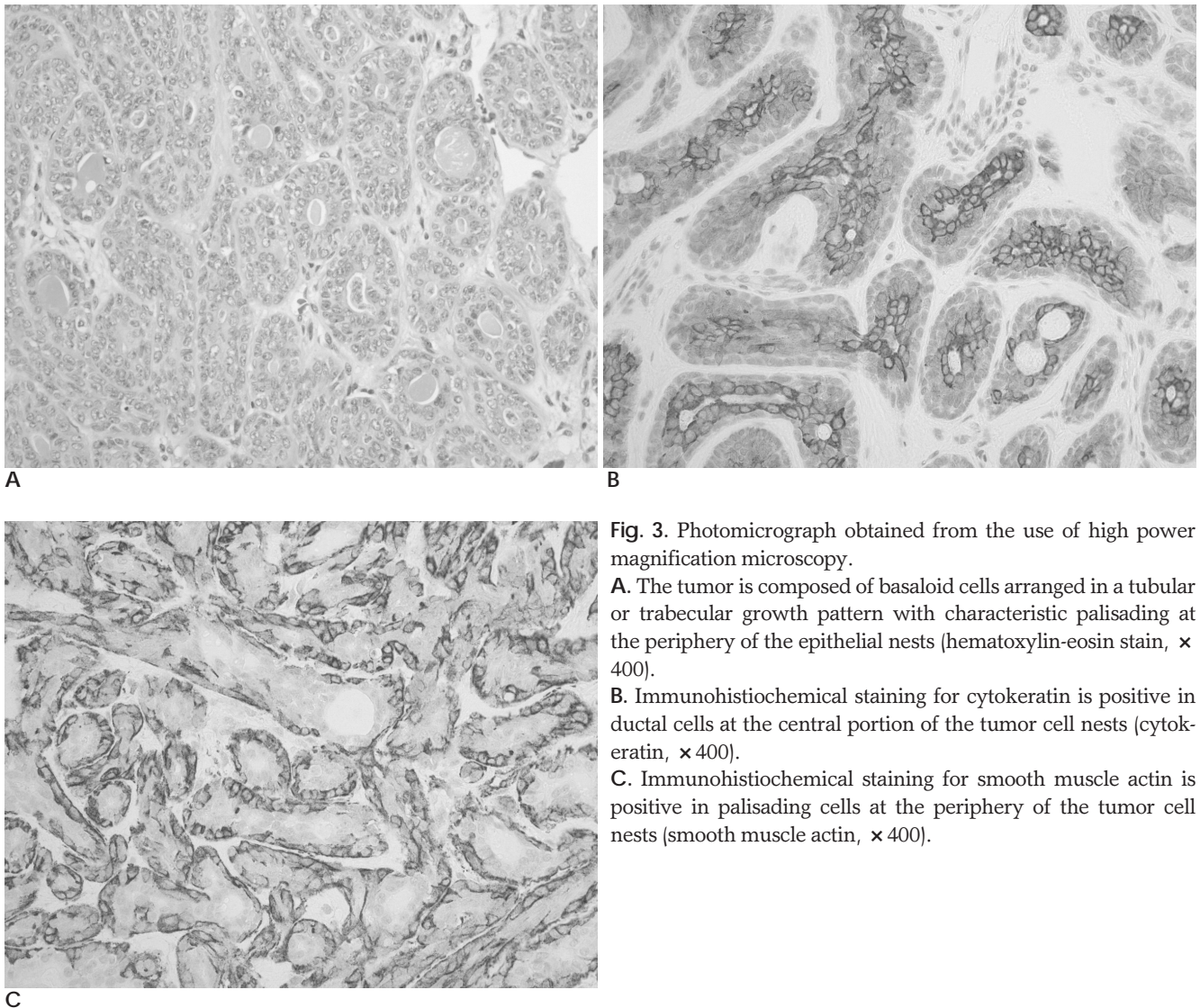


Fig. 3. Photomicrograph obtained from the use of high power magnification microscopy.

A. The tumor is composed of basaloid cells arranged in a tubular or trabecular growth pattern with characteristic palisading at the periphery of the epithelial nests (hematoxylin-eosin stain, $\times 400$).

B. Immunohistochemical staining for cytokeratin is positive in ductal cells at the central portion of the tumor cell nests (cytokeratin, $\times 400$).

C. Immunohistochemical staining for smooth muscle actin is positive in palisading cells at the periphery of the tumor cell nests (smooth muscle actin, $\times 400$).

ed from the stroma by a basement membrane. There is characteristic palisading in the peripheral portion of the tumor cell nests. A myxochondroid stromal component, as seen in pleomorphic adenoma or a mesenchymal component should be absent. It is also positive for cytokeratin, smooth muscle actin, and S-100 protein by immunohistochemical staining (2, 4, 6).

Basal cell adenoma can be divided into four subtypes based on their morphologic pattern: trabecular, solid, tubular, and membranous, and the most frequent type is the solid variant (6, 7). Malignant transformation of basal cell adenoma is rare but has been suggested by some researchers, giving rise to basaloid cell carcinomas (basal cell adenocarcinoma, adenoid cystic carcinoma) and, less commonly, non-basaloid carcinoma (salivary duct carcinoma, adenocarcinoma) (1, 8).

Basal cell adenoma typically appears as a solitary,

slowly growing, asymptomatic mass usually in the parotid gland or the minor salivary gland of the other parts. Surgical extirpation of the tumor has been curative. Although recurrence is rare, the membranous subtype, which is a hereditary variety of basal cell adenoma, has been reported to have a 25 - 37% recurrence rate in some reports (1 - 3, 8).

Imaging features of basal cell adenomas are rarely reported because of their low prevalence. General imaging findings on previously reported cases have demonstrated that these tumors occur mainly in the parotid gland and have well-demarcated margins, solid or cystic components, and homogeneous or heterogeneous enhancement. These imaging findings are not characteristic of a basal cell adenoma.

Grossly, basal cell adenoma is solid or cystic, or gray or white and does not usually show intratumoral hemor-

rhage or calcification. However, Jeong et al. (3) reported that basal cell adenoma arising in the parapharyngeal space shows intratumoral hemorrhage on MR imaging. Basal cell adenoma has a characteristic vascular pattern in which small capillaries and venules are prominent in the adenoma, and these vascular structures can cause intratumoral hemorrhage (3, 9). Chawla et al. (1) have described CT features of basal cell adenomas arising in the parotid glands that correlated with histopathological findings. The tumor showed heterogeneous enhancement on contrast-enhanced CT in 11 of 14 cases (79%), with stellate-shaped low-density areas in three tumors, linear non-enhancing bands in three tumors and cystic areas in five tumors. In our case, the central low-density area in the tumor was seen on contrast-enhanced CT but we could not verify the hypodense area in the tumor pathologically. Some reports have demonstrated the hypointense rim of the tumor capsule on T2-weighted MR images due to tumor encapsulated by fibrous connective tissue (2, 3).

The parapharyngeal space contains mainly fatty tissue, lymphatics, and minor salivary gland tissue, and is actually a potential space located lateral to the upper pharynx and extends from the skull base to the hyoid bone. This space may be the source of salivary gland tumors, as it was in this case (3). Pleomorphic adenoma originated from the minor salivary gland is the most common primary lesion arising in the parapharyngeal space. Basal cell adenoma occurs in the parapharyngeal space very rarely. According to our own investigation, only three cases have been reported (3 - 5).

The major differential diagnosis for parapharyngeal tumors includes other tumors of minor salivary gland origin, including the pleomorphic adenoma, Warthin tumor and other low-grade malignant tumors, nerve

sheath tumors, and paragangliomas. It is difficult to make a differential diagnosis from other minor salivary gland tumors in the parapharyngeal space. Schwannoma or paraganglioma is usually located in the carotid space; therefore, it displaces the internal carotid artery anteromedially. Paraganglioma is a hypervascular tumor and is characterized by flow signal void within the mass on MR imaging.

In conclusion, basal cell adenoma is a rare benign epithelial neoplasm originating from salivary gland tissue and we report here on a very rare case of a basal cell adenoma arising in the parapharyngeal space. Although it has low prevalency and no specific imaging features, we think that it should be considered in the differential diagnosis of parapharyngeal space tumors or salivary gland origin tumors.

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