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Basal Cell Adenoma of the Submandibular Gland

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Abstract: Basal cell adenoma is a benign tumor that appears to have unique histological characteristics and occurs only rarely in the submandibular and sublingual glands. Basal cell adenomas are slowly growing, firm, and asymptomatic masses that are frequently diagnosed during the fifth and sixth decades of life. Aspiration biopsy is important for a correct diagnosis. Submandibular gland excision should be performed for treatment. Although rare, basal cell adenoma should be kept in mind as a differential diagnosis of submandibular gland masses. We herein report a case involving a 39-year-old female with a basal cell adenoma of the submandibular gland.
Keywords: Adenoma; Basal Cell; Submandibular Gland; Benign; Salivary Gland.

INTRODUCTION

Salivary gland tumors are relatively rare, constituting 3% to 4% of all head and neck neoplasms [1]. Approximately 22% of salivary gland tumors arise in the submandibular gland, but most (70%) arise from the parotid gland [1]. Spiro reviewed all salivary neoplasms that occurred within a 35-year in Memorial Sloan-Kettering Hospital and reported that benign neoplasms constituted 54% of all tumors while basal cell adenomas constituted 0.2% [2]. However, the incidence of basal cell adenoma is 1% to 2% of all salivary gland tumors [3-5]. Basal cell adenoma is a benign tumor and is classified by the World Health Organization as an epithelial tumor [6]. Most basal cell adenomas occur in the parotid gland and minor salivary glands of the upper lip [1, 3-5]. Basal cell adenoma occurs only rarely in the submandibular and sublingual glands [3, 5-7]. It is a slowly growing, firm, asymptomatic mass that is frequently diagnosed during the fifth and sixth decades of life [1, 3, 5]. The diagnosis is typically achieved by fine needle aspiration biopsy (FNAB) [3, 4], and the adenoma is histopathologically graded as solid, trabecular, tubular, or membranous [1, 3]. The monomorphic basaloid appearance of the cells is critical in the diagnosis of basal cell adenoma [3-7]. This tumor is a specific type of monomorphic salivary gland tumor.

This case report describes a 39-year-old female with a basal cell adenoma in the submandibular salivary gland, a rare location. The histopathological and clinical features, differential diagnosis, and management are emphasized.

CASE REPORT

A 39-year-old female presented to our hospital with a 4-year history of a painful mass in her left submandibular region. The mass had exhibited intermittent growth and regression during those 4 years. It began as a small mass that progressed to the present size. Physical examination revealed a 20 X 10 mm moderately firm, swollen mass that was tender on palpation. No enlarged lymph nodes or other masses were observed in the head and neck region. Ultrasonography revealed a 12X8mm mass within the left submandibular gland.

A tru-cut biopsy was performed, revealing a diagnosis of basal cell adenoma. An excisional biopsy was then recommended. Tru-cut biopsy examination revealed monomorphic cells with hyperchromatic nuclei, scant cytoplasm, and indistinct nucleoli arranged in a solid and trabecular pattern (Fig. 1). No nuclear atypia, mitosis, or necrosis was noted. The patient underwent total excision of the submandibular salivary gland under general anesthesia (Fig. 2).

The mass was 40 X 30 X 25 mm in size on macroscopic examination of the excised material. An 8 mm diameter, well-circumscribed, encapsulated nodular lesion was observed in the submandibular gland sections (Fig. 3). The cross-sectional aspect was grayish white and solid, and the sections of the lesion were homogeneous in structure. Microscopic examination of the tumor specimen revealed oval nuclei, narrow eosinophilic cytoplasm, cell boundaries that could not

be clearly distinguished, and solid nests of basaloid cells, and a trabecular form. Dense collagenous stroma was observed between the islands (Fig. 4). No mesenchymal component, myxoid matrix, or cystic changes were observed (Fig. 5). The tumor was fully encapsulated and the margins were limited. The cytokeratin 7 (CK7), p63, and smooth muscle actin (SMA) immunohistochemical staining results supported ductal and myoepithelial differentiation. The Ki67 proliferation index was 1% to 2%. Based on these findings, the tumor was diagnosed as a basal cell adenoma. The postoperative course was uneventful. Follow-up was performed for 12 months with no signs of recurrence.

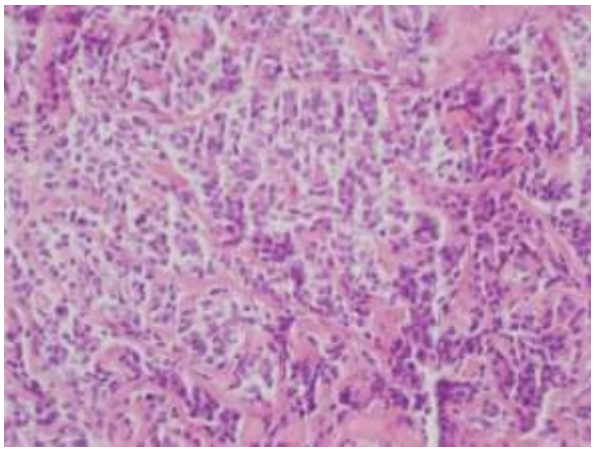

Fig. 1: Monomorphic basaloid cells with hyperchromatic nuclei in tumor tissue (X400 magnification, hematoxylin and eosin staining)

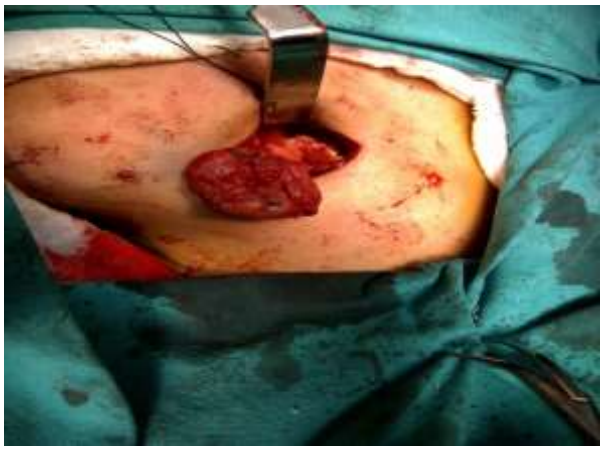

Fig. 2: Surgical excision of the mass

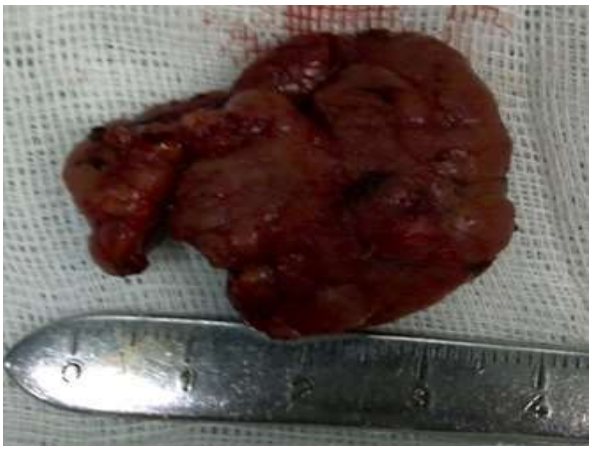

Fig. 3: The mass was 40 X 30 X 25 mm in size

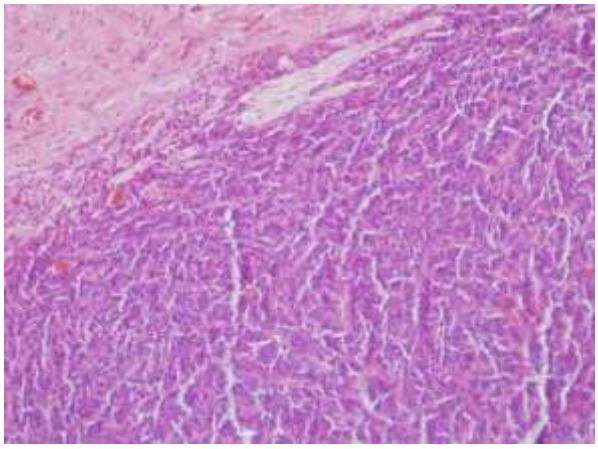

Fig. 4: The solid tumor tissue with dense collagenous stroma and a trabecular pattern (X200 magnification, hematoxylin and eosin staining)

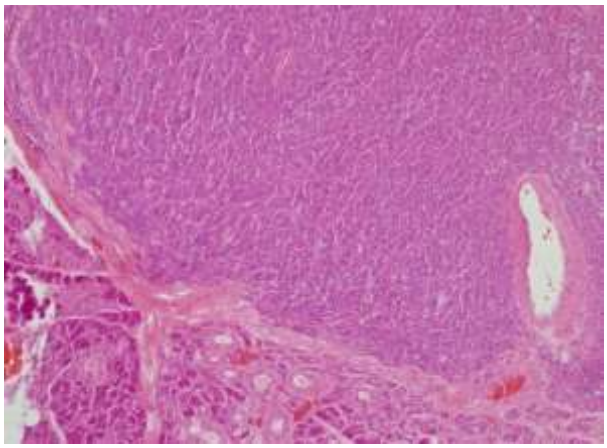

Fig. 5: The tumoral tissue was separated by a fibrous capsule (X100 magnification, hematoxylin and eosin staining)

DISCUSSION

The incidence of basal cell adenoma among all salivary gland tumors is 1% to 2% [3-5]. Kleinsasser and Klein [6] first defined this tumor in 1967, and it was classified as a benign epithelial tumor of the

salivary glands by the World Health Organization in 1991 [7]. The majority of these tumors arise in the parotid gland (70%); they are rare in the submandibular gland and minor salivary glands [1, 3, 4, 8, 9]. Luksic *et al*. reported a series of 779 tumors of the salivary gland, and basal cell adenoma was identified in 6 patients. However, they determined that the basal cell adenomas were located in the parotid gland in all six patients; no patient exhibited a submandibular localization [10]. Kızıl *et al*. evaluated the demographic and clinicopathological data of 510 salivary gland tumors in Turkey. They reported only four basal cell adenomas, all of which were located in the parotid gland; none were found in the submandibular gland [11]. Submandibular localization of basal cell adenomas is very rare, and there are few case reports in the English-language literature [3, 5]. The adenoma was located in the submandibular gland in the present case.

Basal cell adenoma is often seen from the fifth to seventh decades of life [3-5]. Gürbüzel *et al*. [3] and Veeresh *et al*. [5] each reported a case of basal cell adenoma of the submandibular gland, and both patients were 65 years old. However, our patient was 39 years old; both the age of our patient and her particular localization are rare. Basal cell adenomas are more common in females, however [3, 4]; our patient was also female.

Basal cell adenoma grows slowly and presents as a firm, mobile mass [3, 5, 12]. It is usually asymptomatic. The tumor is typically detected when it is ≥3 cm in size [10]. In our case, the tumor was a slowly growing, firm mass that was tender of palpation and that measured 12 X 8 mm.

The diagnosis of basal cell adenoma must be established histologically. Biopsy is generally accepted to be the most accurate method by which to obtain a diagnosis, although some authors advocate FNAB if physical access to the tumor is impossible [5]. We performed a tru-cut biopsy for diagnosis in the present case.

Histopathologically, basal cell adenomas are classified as solid, trabecular, tubular, or membranous. The cells have a monomorphic, basaloid histologic appearance without a chondromyxoidstroma [1, 3]. The most common type is the solid variant [3, 10], as in the present case.

CK is an immunomarker of epithelial cells, while SMA is a marker of myoepithelial cells. These markers show epithelial and glandular origins [3, 5]. Basal cell adenoma is immunoreactive for p63, especially at the periphery, where the tumor cells lie adjacent to the connective stroma [3, 5]. In our case, similar to previous reports, CK7, p63, and SMA immunostaining demonstrated the origin and nature of the cell types.

All clinically asymptomatic and mobile salivary gland tumors should be considered as a differential diagnosis. Thus, it is sometimes necessary to perform complete excision of the tumor prior to making the final diagnosis.

Pleomorphic adenoma is the most frequently encountered tumor in this region, so it should be first considered as a differential diagnosis. Because the clinical features are similar, a histopathological differential diagnosis is important. The absence of myoepithelial cells, an intact basal membrane, and a sharp demarcation between the epithelium and stroma are important findings to differentiate these two tumors from each other [1, 8]. In our case, pleomorphic adenoma was excluded because in addition to the above reasons, the lesion comprised monotonous basaloid cells but no chondromyxoid stromal features.

The differential diagnoses must include some unfavorable entities such as basal cell adenocarcinoma, adenoid cystic carcinoma, and basaloid squamous cell carcinoma. In our case, because of the favorable limited margins; absence of a capsule; lack of vascular and perineural invasion; and absence of necrosis, mitosis, and cellular pleomorphism, we were able to exclude basal cell adenocarcinoma, polymorphous low-grade adenocarcinoma, and other malignant salivary gland tumors as differential diagnoses.

The mainstay of treatment of these benign tumors is surgical excision with a cuff of normal surrounding tissue [1]. Our patient underwent total excision of the submandibular salivary gland, and no tumor tissue was observed within the surgical margins.

The recurrence rate for solid and trabecular-tubular variants is almost 0%. This contrasts with the high recurrence rate (24%) of the membranous type, which is perhaps a result of the multicentricity of this lesion. Although exceedingly rare, malignant transformation is more common in the membranous type than in the other types [1, 3-5]. In our case, recurrence was not observed throughout the 12-month follow-up period.

CONCLUSION

Basal cell adenoma is a rare, benign salivary gland tumor, and localization in the submandibular gland is even rarer. Although it often occurs from the fifth to seventh decades of life, it may also be seen in the fourth decade. FNAB is important for a correct diagnosis. Submandibular gland excision should be performed for treatment. Although rare, basal cell

adenoma should be kept in mind as a differential diagnosis of submandibular gland masses.

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