

SHORT COMMUNICATION

Basal cell adenoma in a relatively rare site

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ABSTRACT

Basal cell adenoma (BCA) of the salivary glands is an uncommon type of monomorphic adenoma. Its most frequent location is the parotid gland. It usually appears as a firm and mobile slow-growing mass. Histologically, it is seen as nests of isomorphic cells and interlaced trabeculae with a prominent basal membrane. There is also slack, hyaline stroma with absence of a myxoid or chondroid component. We describe a case of BCA of palatal minor salivary glands, a rare site for its occurrence. We also briefly review the literature on the same.

Key words: Basal cell adenoma, monomorphic adenoma, squamous morules, pancytokeratin, alpha smooth muscle actin

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INTRODUCTION

The origin of tumors of minor salivary glands account for less than 25% of all salivary neoplasms with 40–80% occurring in the palate.^[1,2] In 1967, Kleinsasser and Klein introduced the term “basal cell adenoma” to describe a benign salivary gland tumor comprised of uniform appearing basaloid cells arranged in solid, trabecular, tubular, and membranous patterns, but lacking the myxoid and chondroid mesenchymal-like component as seen in pleomorphic adenoma.^[3] It has been recognized as one of the nine subcategories of salivary gland adenomas in the Second Edition of the Salivary Gland Tumors Classification of the World Health Organization (WHO), and accounts for approximately 1–2% of all salivary gland epithelial tumors.^[3] It is considered as a low-grade malignant tumor with a high recurrence rate.^[4] It is also called as Monomorphic Adenoma.^[5]

CASE REPORT

A 32-year-old male patient reported with a chief complaint of painless swelling on the right side of the palate, which was first noted by a local physician two months prior. The general clinical examination was otherwise unremarkable.

Examination

On intraoral examination an, asymptomatic solitary swelling with an insidious onset and growth was seen on the palate. On inspection it was ovoid in shape, measuring about 3.5 × 2 cm, with well-circumscribed borders. It extended from the mesial aspect of 14 to the distal aspect of 17, and laterally from the free gingival margin in relation to 14–17 to 0.5 cms away from the midline. The mucosa over the swelling was pinkish-red in color and stretched. There was blanching noted in some areas

over the swelling. On palpation the swelling was nontender, with a smooth surface, and was soft-to-firm in consistency. The mucosa over the swelling was freely mobile. No other lymph nodes or masses were palpable in the head and neck region. An intraoral periapical radiograph and an occlusal radiograph were obtained for the same. A well-defined radiolucency with distinct margins was noted in relation to 14–17. With a clinical diagnosis of adenoma of minor salivary glands, fine needle aspiration cytology was performed.

Investigations

The cytosmears stained with Rapid Papanicolaou stain revealed predominantly basaloid cells arranged in clumps with hyperchromatic nuclei and an increased nuclear-cytoplasmic ratio [Figure 1]. Suspecting it to be an adenoma of minor salivary glands, excisional biopsy was recommended. The biopsy specimen obtained was about 3.0 × 2.0 × 1.5 cms in dimension, brownish-white in color [Figure 2], and soft-to-firm in consistency. It was routinely processed and stained with Hematoxylin and Eosin stain. In addition few sections were stained with Periodic acid Schiff's stain. The sections revealed an overlying orthokeratinized type of stratified squamous epithelium with an underlying connective tissue. In the deeper layers of the mucosa was a well-circumscribed encapsulated tumor mass enclosing multiple islands and cords of epithelial cells, supported by a small amount of fibrous stroma [Figure 3]. The basaloid cells making up the bulk of the tumor were found to be isomorphic. The peripheral cells were palisaded with a cuboidal-to-columnar shape, while the central cells were relatively rounded. These peripheral cells were hyperchromatic, while the central cells had pale staining nuclei [Figure 4]. The sharp demarcation between the neoplastic epithelial cells and the surrounding connective tissue was evident. PAS positive membrane-

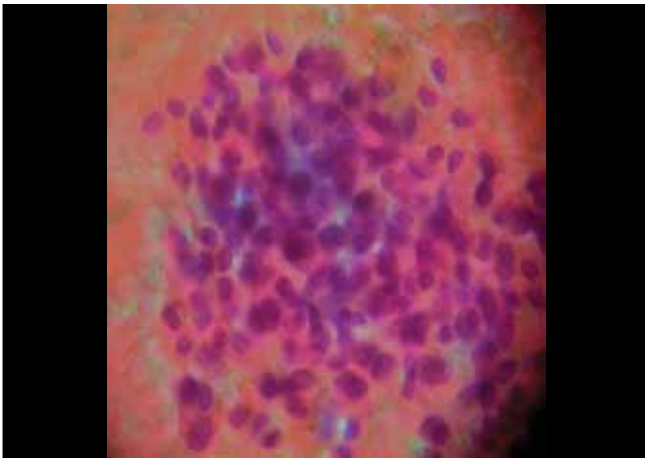


Figure 1: Papanicolaou stained cytosmears with basaloid cells arranged in clumps with hyperchromatic nuclei and an increased nuclear-cytoplasmic ratio



Figure 2: Submitted biopsy specimen, brownish-white in color

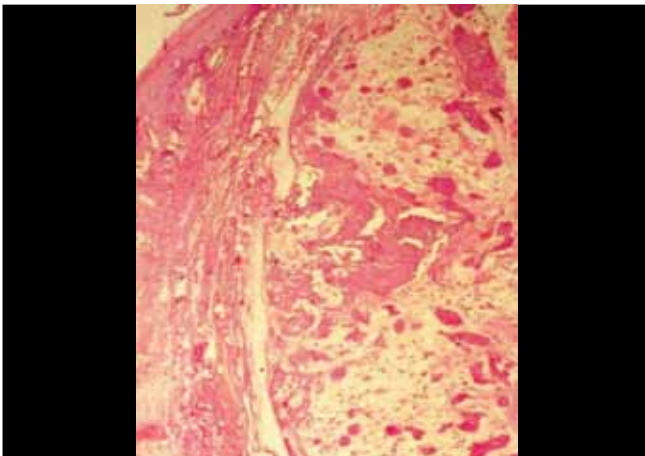


Figure 3: An overlying orthokeratinized type of stratified squamous epithelium with underlying well circumscribed encapsulated tumor mass in connective tissue enclosing the multiple islands and cords of epithelial cells supported by a small amount of fibrous stroma

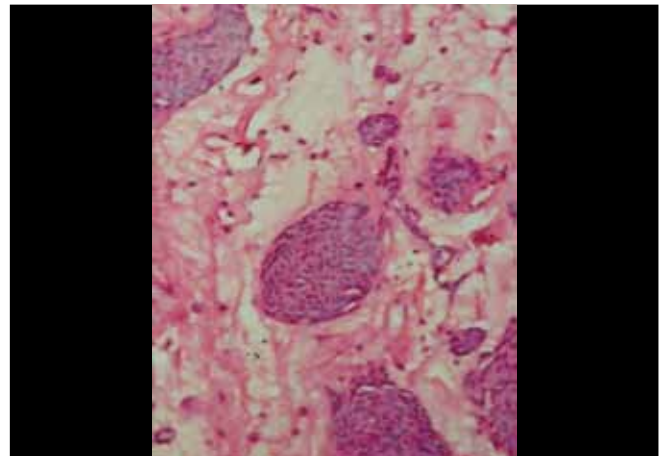


Figure 4: An island of isomorphic cells with peripheral palisaded cuboidal to columnar shape and central cells relatively rounded

like structures surrounding cellular nests showed intense eosinophilia that clearly delimited the basaloid cells [Figure 5]. The above-mentioned features were suggestive of basal cell adenoma.

To confirm our diagnosis, we performed an immunohistochemical examination using antibodies against the markers pancytokeratin and alpha smooth muscle actin. The tumor tissue revealed no positivity for alpha smooth muscle actin [Figure 6], though it was found to be positive in normal salivary gland acini [Figure 7]. The tumor tissue exhibited focal areas of positivity for pancytokeratin [Figure 8]. This confirmed our diagnosis of basal cell adenoma. A follow-up of nine months revealed no recurrences.

DISCUSSION

Basal cell adenoma represents 54% of monomorphous

adenomas and 1 – 3% of major salivary glands tumors with an incidence of 7.5% among primary epithelial parotid gland tumors. It usually affects older persons over 50 years (mean - 57.7 years) with female predilection (1:1.02).^[2,3,6]

They are most often located in the parotid gland (superficial lobe).^[6] The minor salivary gland is a relatively rare location for this tumor.^[7] Our case was in the minor salivary glands of the palate, a rarer site. Most BCA of the parotid gland are unilateral, round or oval in shape with well-circumscribed borders.^[6] Katsuno *et al.* described a rare case of bilateral basal cell adenomas in the parotid glands of a 65-year-old woman.^[8] A painless mass, enlarging slowly is the most common clinical symptom.^[6] Usually there is no regional lymph node involvement, and no calcification or cystic component within the tumor.^[3] It frequently measures between 3 – 8 cm.^[3,4]

Basal cell adenoma may be predominantly solid or

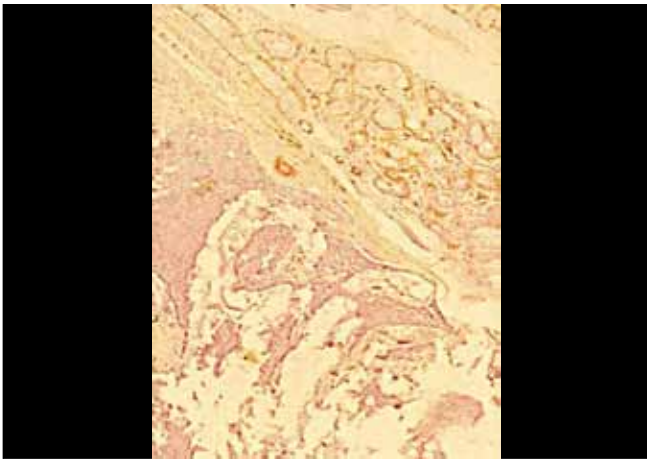


Figure 7: Alpha smooth muscle actin showing positivity only in areas of normal salivary gland acini

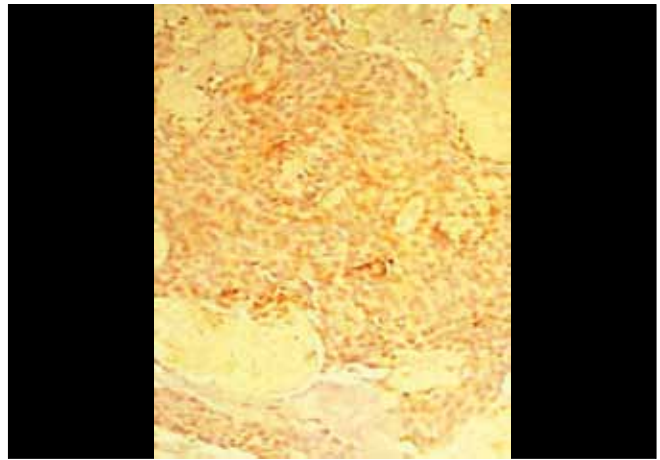


Figure 8: Focal areas of positivity for pancytokeratin

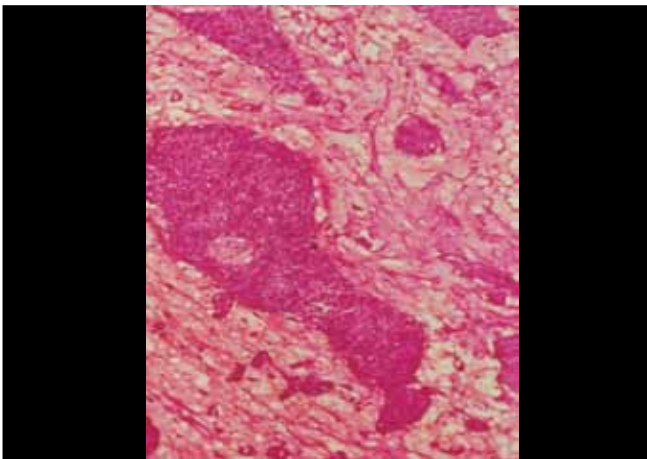


Figure 5: PAS stain clearly exhibiting intensely eosinophilic basal membrane-like structures surrounding cellular nests clearly delimiting basaloid cells

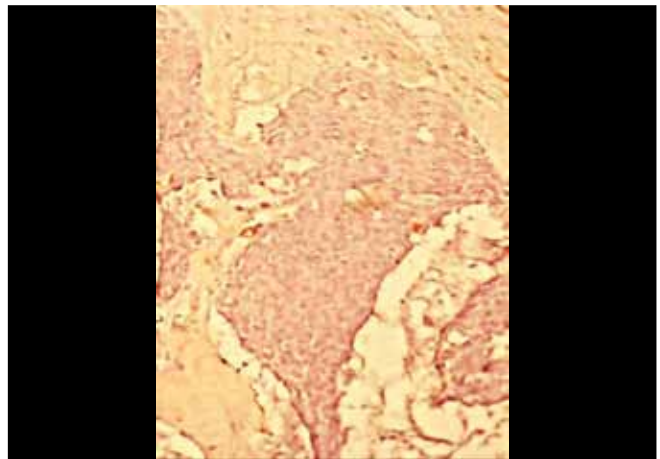


Figure 6: No immunoreactivity for alpha smooth muscle actin

cystic.^[6] The resected mass is usually yellowish-white in color, soft-to-firm in consistency, well-circumscribed, encapsulated and highly vascular.^[6] The color of the cut surface is usually uniform and gray-white or tan-white.^[3]

Fine Needle Aspiration cytology is highly sensitive at detecting basaloid neoplasms.^[9] Aspirates consist of two populations of basaloid cells: a group of small oval cells with bland hyperchromatic nuclei, scant cytoplasm, and indistinct nucleoli, and a group of larger oval-to-polygonal cells, with moderate amounts of delicate pale cytoplasm. The basaloid cells are uniform and haphazardly arranged in clusters of variable size or in trabeculae, often with peripheral palisading of the smaller population of cells. Squamous morules, a characteristic feature of basal cell tumors, are sometimes present in well-sampled cases.^[9]

Histopathologically, these neoplasms feature as relatively uniform, small, dark basaloid epithelial cells in the stroma, and are surrounded by a fibrous connective tissue

capsule.^[6] Palisading at the periphery of the epithelial nests results in a ‘basaloid’ appearance.^[5] The basaloid cells have two different morphologies and are intermingled. One group consists of small cells with little cytoplasm and intensive basaloid rounded nuclei that are usually located in the periphery of the tumoral nests or islands. The other group is formed by large cells with abundant cytoplasm and pale nuclei that are located in the center of the tumoral nests. A basal membrane-like structure surrounds these tumoral nests and clearly separates them from the surrounding nonmucoid stroma. Globally, as it has been referred to in classic texts, the tumor adopts an ameloblastoma-like pattern.^[4] Individual tumors commonly display a combination of several growth patterns.^[3] The basaloid cells are arranged in solid, trabecular, tubular, and membranous pattern.^[6] Ultrastructurally, the tumor appears to be derived from the intercalated portion of the duct, with minor participation of the myoepithelial cells.^[5] Basal cell adenocarcinoma is a rare malignant counterpart of BCA and may be present as a solid, tubulotrabeular, or a membranous subtype.^[9]

Ultrastructural analysis of a case of basal cell adenoma reported by Hemachandran *et al.* showed characteristic features including reduplicated basal laminae around the tumor cells, presence of intermediary filaments, and rough endoplasmic reticulum in the cytoplasm of the tumor cells.^[7]

Immunohistochemistry

Basaloid cells in trabecular and solid areas express vimentin, actin, β subunit of S-100 protein, and cytokeratins. In our case the tumor cells in the islands were focally positive for cytokeratins and negative for alpha smooth muscle actin. Hemachandran *et al.* reported the first case of BCA of the minor salivary gland, which showed selective positivity for pancytokeratin, S-100, and smooth muscle actin in the tumor, which highlighted the participation of myoepithelial cells in histogenesis.^[7]

Clinical differential diagnosis includes mucocele, which usually appears in the lower lip of young people, whereas, BCA appears in the upper lip of the elderly.^[4] Warthin's tumor and membranous BCA are usually unilateral with pleomorphic adenoma and tubular basal cell adenomas exhibiting multicentricity or bilaterality.^[11,12] Among the radiographic differential diagnoses, pleomorphic adenoma and malignant salivary gland tumors mostly show increased enhancement in delayed phases on computed tomography (CT).^[6] The age of the patient and the attenuation on unenhanced and contrast-enhanced CT may help in differentiating BCA from pleomorphic adenoma.^[13,14] Histopathological differential diagnosis includes salivary gland basaloid lesions such as basal cell adenocarcinoma (solid form), benign mixed tumor, adenoid cystic carcinoma, cellular pleomorphic adenoma, chronic sialadenitis, cutaneous basal cell carcinoma, metastatic basaloid squamous carcinoma, and sialoblastoma.^[5,9]

CONCLUSION

Definitive diagnosis by clinical and imaging techniques alone is difficult. The final diagnosis requires histopathological analysis, which can be inverteated by immunohistochemical examination. In any suspected neoplastic salivary gland lesion, due to prognostic implications, differential diagnosis with malignant counterparts is mandatory. This case report describes a case of basal cell adenoma in a

relatively rare site, that is, the hard palate, which had been immunohistochemically confirmed.

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